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**Development and evaluation of an online version of
the *Feeling Better* pain management programme for
children with chronic pain and their care-givers**

Thesis submitted to the National University of Ireland, Galway
in fulfilment of the
requirements for the Degree of Doctor of Philosophy
(Psychology)

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Declaration

I declare that this thesis has not been submitted as an exercise at this or any other university.

I declare that this thesis is entirely my own work.

Signed: _____

Angeline Traynor

Statement of Contribution

The candidate was responsible for leading all aspects this research including the study design, data collection, analysis, interpretation and write-up of each of the four studies that comprise this work. The supervisory team, Graduate Research Committee and relevant experts advised and provided support in conducting the research.

Publications resulting from this thesis

Traynor, A., Morrissey, E., Egan, J., & McGuire, B. E. (2016). The effectiveness of information and communication technology-based psychological interventions for paediatric chronic pain: protocol for a systematic review, meta-analysis and intervention content analysis. *Systematic Reviews*, 5(1), 175. article. <http://doi.org/10.1186/s13643-016-0350-1>

Manuscripts in preparation

Study 1: Chapter 3: Traynor, A, Morrissey, E., Egan, J McGuire, B.E. The effectiveness of information and communication technology-based psychological interventions for paediatric chronic pain: a systematic review, meta-analysis and intervention content analysis.

Study 2: Chapter 4: Traynor, A, O'Higgins, Egan, J., Durand, H. & McGuire, B.E: Living with chronic pain and using technology to support self-management – a participative study.

Study 3: Chapter 5: Traynor, A, Egan, J., Morrissey, E., & McGuire, E: Development of an online treatment platform for pre- adolescent children with chronic pain.

Study 4: Chapter 6: Traynor, A, Egan, J. & McGuire, B.E. Chapter 6: Findings from a pilot randomised control trial of novel, online pain management programme for pre- adolescent children with chronic pain.

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Abstract

Background: Reasons for poor management of paediatric chronic pain include lack of access to psychological support for pain management. Research evidence is increasingly focused on online interventions as a source of treatment delivery. However, research in this field tends to focus on adolescents with chronic pain. There is a gap in our understanding of the feasibility, acceptability and benefits of such interventions for pre-adolescent children with chronic pain.

Aim: The current research focuses on the adaptation and feasibility evaluation of a manual-based pain management programme for pre-adolescent children with chronic pain and their care-givers. It was proposed that a manualised pain management programme called *Feeling Better* could be adapted and delivered remotely to support school-age children (5-12 years) with chronic pain.

Methods: The studies conducted in this research were guided by the UK Medical Research Council Framework for developing complex interventions and a stepped intervention mapping protocol. This involved a review of the evidence base and an appraisal of needs for intervention development, adoption and implementation in four separate but inter-linking studies. In Study 1, a systematic review, meta-analysis and intervention content analysis was conducted to (i) determine the effectiveness of psychological therapies delivered using information and communication technology for children and adolescents with chronic pain and (ii) identify the theoretical basis, behaviour change techniques and modes of delivery that characterise existing interventions and be associated with estimates of effect. The outcomes assessed included pain intensity, pain-related disability, psychological distress (anxiety and depression), quality of life and treatment satisfaction at post-treatment and follow-up among children and adolescents with chronic pain. In Study 2, a participative qualitative study was used to explore the lived experience of chronic pain management and the factors that influence coping behaviour from the perspective of pre-adolescent children with chronic pain and their care-givers. A 3-phase participative protocol was employed to explore

experiences, barriers and facilitators of adaptive coping behaviour with separate groups of children and parents. A theoretical analysis of study findings was undertaken to explore the utility of Social Cognitive Theory as a guiding theoretical framework for intervention development. The sample consisted of 11 children aged 5-12 years, with juvenile idiopathic arthritis and 21 parents recruited from the community using offline and online strategies. In Study 3, a mixed method approach combining quantitative and qualitative methodologies was used to test the usability and acceptability of prototype versions of the adapted *Feeling Better* programme. A participative research process approach was combined with online user testing to explore how children perceive technology-based therapy and what design features are most likely to engage this population. A total of 24 children, aged 5-12 years with juvenile idiopathic arthritis or ehlers-danlos syndrome and 58 care-givers contributed to this mixed method, multi-phased study. The research process involved think-aloud group feedback, a 3-phase participative protocol and online user testing involving online assessment of usability and acceptability. In addition, experts in chronic pain management and intervention development reviewed the prototype and informed subsequent modifications. In Study 4, the feasibility and potential effectiveness of an online version of the *Feeling Better* pain management programme was evaluated in a two-arm, parallel feasibility trial. Eligible participants were randomised using variable block randomisation to one of two groups: (1) the online, cognitive behaviour therapy-based *Feeling Better* intervention or (2) a waitlist control group. The *Feeling Better* intervention was delivered online over a period of 9 weeks. The primary feasibility outcomes were: recruitment, retention, treatment adherence and satisfaction with treatment. The primary clinical outcomes were chronic pain intensity and physical limitations (physical health). Secondary clinical outcomes were mood (psychosocial health), self-efficacy, pain catastrophising, pain coping and parental protectiveness. Assessments were performed at baseline, at post-treatment and at 3-month follow-up.

Findings: The systematic review (Study 1) found tentative evidence to support the effectiveness of ICT-based psychological therapies for

chronic headache pain at post-treatment. Qualitative and quantitative assessment of treatment satisfaction suggests psychological therapy delivered using ICT is acceptable to children and adolescents. Treatment satisfaction was significantly higher in the treatment group compared to the control group. Interventions that excluded the BCT 6.1 ‘demonstration of the behaviour’ and entertainment modes of delivery were associated with greater estimates of effect in response to treatment compared to interventions that were perceived to have incorporated this technique. No significant differences were observed in estimates of effect based on the number or frequency of theoretical constructs, behaviour change techniques or modes of delivery identified. These results should be interpreted with caution as the quality of the included trials was considered low and lack of data prevented several planned analyses. The findings from the participative study (Study 2) suggest children and parents use a range of self-reliant, avoidant, dependent and inconsistent coping approaches. Children and parents differed in the importance they attribute to different pain coping strategies. For children, meaningful activity and goal pursuit strongly influenced engagement in active coping behaviour. For parents, strategies that facilitate emotional equilibrium and communication at home were most important. Seven common categories of barriers (support needs) and facilitators (coping preferences / targets for treatment) were separately identified by children and parents as important influences on coping behaviour and used to inform intervention development. These include i) Being active (physical capability) (ii) Things you love doing (meaningful activities), (iii) Be with Mom (coping habits), (iv) Emotion / relief from emotional distress, (v) Friends and practical help (social support / provision), (vi) Concentration (cognitive capability) and (vii) Find the right help (training). These findings validated the selection of Social Cognitive Theory as a guiding theoretical framework and helped to refine the *Feeling Better* prototype. The findings from mixed-method usability testing (Study 3) indicated the iteratively developed, online *Feeling Better* programme was acceptable and relevant from the perspective of end-users. Suggestions for improvement directly informed each iteration of the prototype website and

implementation of clinical content. This study provided preliminary support for the acceptability, relevance and functionality of the online intervention. In Study 4, the adapted online version of the *Feeling Better* programme was found to be feasible and potentially effective for chronic pain management. The target for recruitment was exceeded. A total of 67 children with chronic pain and their care-givers were recruited. However, attrition at post-treatment was 28% and this was significantly higher at follow-up assessment. The target completion rate of 70% was achieved, a total of 48 (74%) parent-child dyads completed the programme and post-treatment assessment. Treatment satisfaction was high among those in the intervention group. Significant differences were observed between intervention and control groups across all clinical outcomes. Estimates of beneficial effect were medium to large. A significant effect of time was also observed across all outcomes except child-reported psychosocial health and pain intensity. Further feasibility testing is recommended to determine optimal intervention intensity, parental involvement and assessment of outcomes in this population. Overall these findings support the feasibility and potential effectiveness of the online version of the *Feeling Better* programme and a full trial is warranted.

Conclusion: Study 1 suggests psychological therapy delivered using ICT for children with chronic headache has potential and further research is needed. This study characterised the previously unknown theoretical basis and core components incorporated in existing technology-mediated, paediatric chronic pain interventions. The findings of this review offer valuable information for healthcare professionals working in chronic pain services and to researchers involved in designing and evaluating information and communication technology-based interventions. Study 2 offers insight into the subjective experience of chronic pain management in a relatively neglected sub-group of the pain population using a novel research approach. This is the first study to give pre-adolescent children a voice in the development of intervention dedicated to school age children. The children and parents in this study demonstrated a relevant and valuable understanding of the process of effective pain self-management. Study 3

demonstrates the value of usability testing and a person-centred approach to intervention development. Important issues related to programme usability and acceptability were identified and addressed. Study 4 established the previously unknown feasibility and potential effectiveness of online interventions for paediatric chronic pain. A full RCT of the online *Feeling Better* programme was considered feasible and warranted.

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List of Abbreviations

BCT	Behaviour Change Technique
BCTTv1	Behaviour Change Technique Taxonomy version 1
CBT	Cognitive Behavioural Therapy
CORE-Q	Consolidated Criteria for Reporting Qualitative Research
HCPs	Health Care Providers
HRQOL	Health-Related Quality of Life
ICPI	Internet Chronic Pain Intervention
IMPACT in Clinical Trials	Initiative on Methods, Measurement, and Pain Assessment in Clinical Trials
M	Mean
MRC	Medical Research Council
N	Number
NICE	National Institute for Health and Care Excellence
NRS	Numerical Rating Scale
PRISMA	Preferred Reporting Items for Systematic Reviews & Meta
PROSPERO	International Prospective Register of Systematic Reviews
RCT	Randomized Controlled Trial
SD	Standard Deviation
SPSS	Statistical Package for Social Science
TDF	Theoretical Domains Framework
WHO	World Health Organisation
MoD	Mode(s) of delivery

Chapter 1 Introduction

1.1 Chapter overview

The aim of this chapter is to describe the empirical basis for this research and introduce key issues relevant to the overall research project. This chapter is presented in three sections. Section I is concerned with understanding chronic pain and outlines key aspects including classification, prevalence and impact. Section II gives an overview of relevant theoretical models relating to the experience of chronic pain and behaviour change. Section III outlines relevant assessment and treatments options supporting chronic pain management including remotely delivered psychological therapies and the development of complex interventions. This chapter concludes with an overview of the structure of this doctoral thesis including a brief rationale for this research based on the limitations of the existing evidence base and the aims and objectives of each study supporting this work.

1.2 Section I: Understanding chronic pain

1.2.1 Definition and classification of chronic pain

Empirical evidence suggests the mechanisms underlying chronic pain are physiological, emotional, cognitive, and environmental (Turk, Wilson, & Cahana, 2011). Thus, the International Association for the Study of Pain defined pain as, "*an unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage*" (Merskey & Bogduk, 1994, p 210-211). In other words, pain is a complex, subjective experience which is unique to the individual.

The complex and subjective nature of the pain complicates assessment. Pain is typically classified in terms of its intensity (acute, chronic), pathophysiology (neuropathic, nociceptive or mixed) or type (arthritic, myofascial, malignant or diabetic). Acute pain is pain resulting from injury, surgery or trauma, which lasts for a short period and diminishes as healing occurs. Chronic pain is persistent or recurrent (episodic) pain that

is resistant to the natural course of healing (Palermo et al., 2012; Friedrichsdorf et al., 2016).

Chronic pain may be described as pain that persists for a period of three to six months or more, however it is more acceptable to define chronic pain in terms of functional rather than arbitrary, temporal parameters (Turk & Okifuji, 2002). Chronic pain may present as a primary pain disorder (e.g. headache, musculoskeletal or abdominal pain) or the primary symptom of an underlying condition (e.g. ehlers-danlos syndrome). Some phenotypes may be experienced simultaneously (i.e. multiple or widespread pain). Pain may be nociceptive (i.e. associated with injury or damage to the body including surgery) such as low back pain or neuropathic (i.e. associated with damage to or dysfunction of the nervous system) such as carpal tunnel syndrome or complex regional pain syndrome (Merskey & Bogduk, 1994; Jensen et al., 2011; IASP, 2012). The complex nature of the chronic pain and issues related to pain assessment may have contributed to a reported under-treatment and under-recognition of chronic pain in young people, particularly younger children and those with primary pain disorders (Jensen & Karoly, 1991; Miró, Huguet, & Jensen, 2014; Friedrichsdorf et al., 2016).

1.2.2 Epidemiology of chronic pain

Chronic pain is a significant problem among adults (Breivik, Collett, Ventafridda, Cohen, & Gallacher, 2006; Hoy, Brooks, Blyth & Buchbinder, 2010; Raftery et al., 2011), adolescents and children (Palermo & Chambers, 2005; Jeffries, Milanese, & Grimmer-Somers, 2007; Gieteling, Bierma-Zeinstra, Passchier, & Berger, 2008; King et al., 2011; Swain et al., 2014). Conservative prevalence estimates suggest that between 11% and 38% of children across Western countries experience chronic pain lasting for a period of three months or longer (Perquin et al., 2000; Stanford, Chambers, Biesanz, & Chen, 2008; King et al., 2011). The most commonly reported paediatric chronic pain conditions are musculoskeletal pain, abdominal pain and headache (King et al., 2011; Palermo et al., 2012). With a median prevalence rate of up to 23%, headache is the most commonly studied pain

condition among children and adolescents (King et al., 2011). Prevalence estimates are influenced by factors such as gender, age and socioeconomic status. Chronic pain is associated with higher prevalence rates among girls and those from lower socioeconomic backgrounds. Moreover, the trajectory of chronic pain has been shown to increase with age (Hunfeld, Perquin, Bertina, Hazebroek-Kampschreur, van Suijlekom-Smit, Koes, & Passchier, 2002). These estimates suggest paediatric chronic pain is a significant healthcare problem.

1.2.3 Impact of chronic pain

Chronic pain is associated with a wider range of co-morbidities than possibly any other condition. These co-morbidities can have a modifying effect on the clinical presentation of pain and must be a considered part of the assessment and treatment approach. There is wealth of empirical research showing that the impact of paediatric chronic pain is pervasive across every domain of functioning and over time. Research shows that untreated chronic pain is predictive of persistent pain or psychological disorder later in life (Mulvaney et al., 2006; Dunn et al., 2011; Burke, Finn, McGuire, & Roche, 2017).

Although the majority of children who present with chronic pain are not severely disabled by their condition, a significant minority, approximately 5–15 %, report severe levels of pain requiring extensive and costly rehabilitation (Huguet & Miró, 2008; Hechler et al., 2015). For others, pain intensity impedes daily functioning (Palermo, Wilson, Peters, Lewandowski, & Somhegyi, 2009).

Disablement associated with chronic pain is often exacerbated by psychological factors. Children coping with long term pain are at greater risk for internalizing problems (Mulvaney, Lambert, Garber & Walker, 2006; Dunn, Jordan, Mancl, Drangsholt, & Le Resche, 2011). Long term, moderate to severe pain is consistently associated with co-morbid conditions such as anxiety and depression (Jacobson et al., 2013; Kashikar-

Zuck, Zafar, et al., 2013). A recent systematic review of sex differences in the efficacy of psychological therapies for the management of chronic pain found that girls report more psychological distress (e.g. anxiety and depression) than boys (Boerner, Eccleston, Chambers, & Keogh, 2016). Further, anxiety was related to poorer outcomes in response to cognitive behavioural treatment for paediatric pain (Cunningham et al., 2016).

Long term pain is associated with sleep disturbance (Meltzer, Logan, & Mindell, 2005; Lewandowski, Ward, & Palermo, 2011; De la Vega & Miró, 2012; Palermo et al., 2009; Shyen et al., 2013), social (Forgeron et al., 2010, Forgeron et al., 2011) and family functioning problems (Palermo & Chambers, 2005; Lewandowski, Palermo, Stinson, Handley, & Chambers, 2010; Jaaniste, Phipps, Lang, & Champion, 2013).

For parents, the negative effects are exacerbated by an associated economic burden resulting from loss of income to facilitate childcare and healthcare utilization (Sleed, Eccleston, Beecham, Knapp, & Jordan, 2005; Phillips, 2006; Palermo et al., 2012; Groenewald, Essner, Wright, Fesinmeyer, & Palermo, 2014). The economic burden associated with chronic pain in adulthood is also significant (Raftery et al., 2012). This is a significant issue as longitudinal studies have observed that approximately 17% of adults with chronic pain report the experience of chronic pain in childhood. In one study, approximately 80% of adults reported that their childhood pain was ongoing (Hassett et al., 2013).

The increasing impact of chronic pain suggests that early intervention is indicated, particularly for girls to prevent the development of significant psychological distress (Boerner et al., 2016). Also that the development of effective, accessible interventions should be a key area of concern for health research and care (Raftery et al., 2012; Sturgeon, 2014). Health research may be guided by consideration of the conceptual and theoretical models that attempt to understand and explain chronic pain.

1.3 Section II: Conceptual and theoretical considerations

Conceptual and theoretical considerations are summarised below with specific focus on the more comprehensive Biopsychosocial Model (Engel, 1977) and Fear-avoidance model (Lethem et al., 1983; Vlaeyen, Kole-Snijders, Rotteveel, Ruesink, & Heuts, 1995). Aspects of each model informed the development of studies within the current thesis and are summarised below.

1.3.1 Biomedical model of chronic pain

The traditional Biomedical Model explains pain in terms of tissue damage and disease postulating that the greater the damage the greater the pain intensity (Keefe et al., 2005). From this perspective, when the organic cause of pain is cured the experience of pain will dissipate. In the absence of an organic cause, pain is labelled psychogenic or arising from psychological factors and therefore ‘not real’ (Gatchel, 2004). This understanding of chronic pain is too simple. The biomedical perspective fails to capture the complexity of the chronic pain experience because it considers only the biological variables associated with disease and not the psychosocial and behavioural variables of illness (Engel, 1977). It cannot explain why the clinical presentation of chronic pain (e.g. pain intensity and associated disability) does not always correspond to the physical pathology (Turk & Okifuji, 2002; Keefe & Somers, 2010).

Gate Control Theory (GCT) put forward by Melzack and Wall (1965;1996) was the first integrated model of physiological and psychological processing that adequately accounts for the varied aspects of the pain experience. According to GCT, the brain is an active recipient of nociceptive stimuli and plays a key role in the interpretation of pain signals. This process is further influenced by psychological and sensory components (Katz & Rosenbloom, 2015). Psychological components include the amplifying effects of emotion and the interpretative role of cognition in the experience of chronic pain (Gatchel, Peng, Peters, Fuchs, & Turk, 2007). This recognition of the complexity of the pain experience laid the foundation for a biopsychosocial understanding for chronic pain. The

biopsychosocial perspective is the most widely accepted and heuristic account of chronic pain and associated treatment (Gatchel, 2004).

1.3.2 Biopsychosocial model of chronic pain

To develop an evidence-based intervention it is necessary to understand the factors that contribute to adaptive or maladaptive pain behaviour. The biopsychosocial model suggests a combination of biological, psychological and socio-cultural factors influence the experience and expression of pain. This can be seen in Figure 1.1.

The biological part of biopsychosocial is hypothesised in several fields of research, each of which have offered important insight into the aetiological mechanisms of chronic pain. The one commonality across contributions is that it remains unclear if the insights offered are associative or causative. Central sensitization (CS) is an established and evidence-based account of chronic pain. CS is defined as an amplification of neural signalling within the CNS that elicits pain hypersensitivity (Woolf, 2011; Woolf, 2018). Woolf (2011) explains this as increased neural responsiveness to painful and non-painful stimuli. Pain hypersensitivity may be observed in the absence of inflammation or nerve damage and may contribute to the development and chronicity of multiple pain phenotypes (Woolf, 2011; Friedrichsdorf et al., 2016). CS is an account of chronic pain, credited with providing a mechanistic explanation with which to understand “unexplained” clinical pain conditions and thereby offer a therapeutic target (Woolf, 2011; Palermo et al., 2012). The HPA axis and stress regulation system may also contribute to the pathology of chronic pain (Melzack, 2001; 2005). This account posits that the experience of chronic pain is a stressor causing prolonged activation of the stress regulation system. Over time this leads to the breakdown of muscle and neural tissue that can in turn, cause pain and promote a cycle of pain–stress–reactivity (Gatchel et al., 2007). This account is supported by empirical evidence which shows a relationship between cortisol secretions (i.e. a measure of the stress response) and the development of chronic pain (Gatchel et al., 2007). Brain

imaging, and neuroscience research have also made important discoveries in understanding the structural, neural and biochemical mechanisms involved in pain processing. This has led to important clinical applications such as the development of analgesic agents for managing pain and the identification of common pathogenic mechanisms and neurotransmitters involved in chronic pain and mood disorders such as depression. Finally, new areas of research include the study of genetic factors which might offer insight into the aetiological mechanism of pain and individual differences in the pain experience and treatment response.

The psychosocial part of the biopsychosocial model refers to the psychological (e.g. cognitions, emotions) and socio-cultural influences (e.g. age, education, ethnicity, social norms) that shape the pain experience. The causal mechanisms underpinning concurrent association between psychosocial factors remain unclear. To develop an effective therapy-based intervention it is important to understand these factors at a theoretical and practical level.

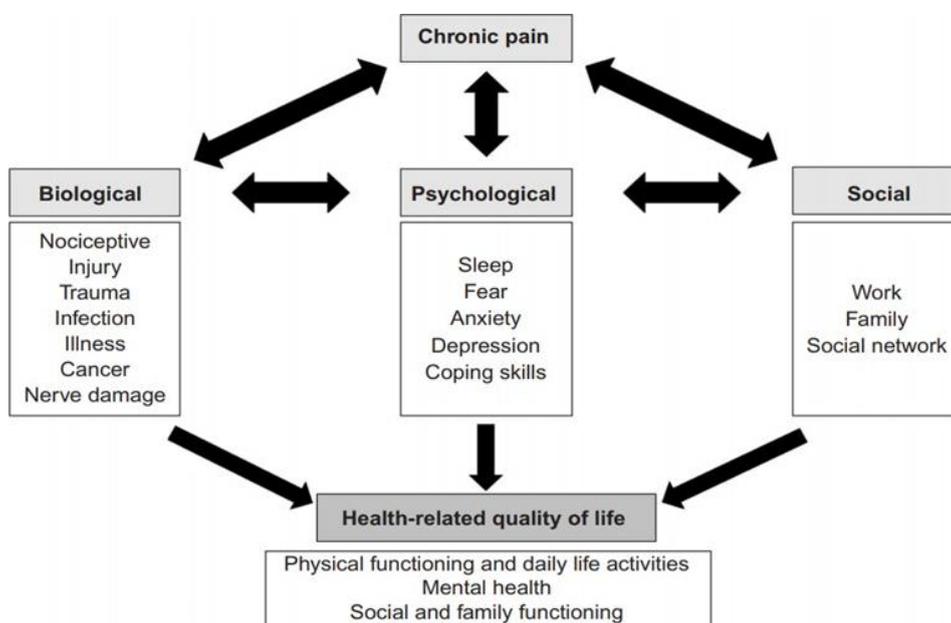


Figure 1.1 Biopsychosocial model of pain and consequences on quality of life (Dueñas, Ojeda, Salazar, Mico, & Failde, 2016; <https://doi.org/10.2147/JPR.S105892> [CC BY-NC 3.0]).

1.3.3 Beliefs and appraisals

Pain beliefs are assumptions about reality that shape how the individual interprets painful events. They are developed over time, based on experience and considered key determinants of pain appraisal i.e. the meaning attributed to pain (Van Rysewyk, 2016). Correlational research provides preliminary evidence that patient beliefs and appraisal of pain events shape the experience of pain and subsequent pain adjustment (Jensen & Karoly, 1991; Miró et al., 2014). This research shows, beliefs about ability to control pain (e.g. self- efficacy; SE), what pain means (e.g. pain is a signal of inherent damage and disability) and worry about what the future holds (e.g. catastrophising; outcome expectations) affect functioning and quality of life (Walker, Baber, Garber, & Smith, 2008; Kashikar-Zuck, Sil, et al., 2013; Rachel, Robert, Michelle, & Jason, 2014; Wojtowicz, Greenley, Gumidyala, Rosen, & Williams, 2014). Thus, cognitions (beliefs and appraisals) are core determinants of pain related behaviour i.e. affective and behavioural response to pain and important target for psychological treatment (Gatchel et al., 2007; Gatchel, McGeary, McGeary, & Lippe, 2014).

1.3.4 Fear and catastrophising

Catastrophic appraisal of pain events and fear avoidant beliefs are central to a maladaptive response to pain and contribute to its chronicity (Gatchel et al., 2007). Fearful beliefs about pain can lead to the adoption of passive coping strategies e.g. resting or avoiding movement. This is an adaptive response to acute pain but a maladaptive approach to coping with chronic pain. For example, selectively attending to the pain location i.e. hypervigilance can lead to a disproportionate focus on somatic sensations that might otherwise be ignored. In turn this can foster a fear of activities the individual believes might exacerbate pain i.e. fear avoidance. Chronic pain patients with elevated pain-related anxiety anticipate high levels of pain and typically demonstrate poorer cognitive and behavioural response e.g. worry or self- reported disability (George et al., 2010; Cunningham et

al., 2016). Research has shown that fear and anxiety can be more strongly associated with self-reported disability and loss of function than biomedical variables or the characteristics of pain e.g. severity (Turk & Okifuji, 2002; Asmundson, Noel, Petter, & Parkerson, 2012; Crombez, Eccleston, Van Damme, Vlaeyen, & Karoly, 2012).

An overwhelming body of evidence also shows pain catastrophising as a determining factor in adjustment to chronic pain. Catastrophic appraisal of pain events is a tendency toward exaggerated negative thinking about an actual or anticipated pain experience (Sullivan et al., 2001; Gatchel et al., 2007). This pattern of thinking is associated with increased pain, symptom-focused behaviour and dependent coping (i.e. high levels of pain seriousness and low levels of pain coping efficacy) in clinical and non-clinical pain populations (Gatchel et al., 2007; Walker et al., 2008). Pain-related anxiety and catastrophic thinking are consistently associated with reduced physical and psychological functioning and diminished health-related quality of life (Burns, Day, & Thorn, 2012; Leung, 2012; Kashikar-Zuck, et al., 2013; Lynch-Jordan, Kashikar-Zuck, Szabova, & Goldschneider, 2013; Wojtowicz et al., 2014).

1.3.5 Self-efficacy

Self-efficacy (SE) refers to a person's belief in their ability to influence their quality of functioning and perform in ways that afford some control over their experiences (Bandura, 1997). When applied to chronic pain, SE is understood as one's confidence in one's ability to function effectively despite pain (Nicholas, 2007). Greater self-efficacy scores are associated with better physical and psychological functioning and improved overall quality of life in children and adults with chronic pain (Arnstein, Caudill, Mandle, Norris, & Beasley, 1999; Jackson, Wang, Wang, & Fan, 2014; DasMahapatra, Chiauzzi, Pujol, Los, & Trudeau, 2015; Dobson, 2015). Cioffi (1991) posits four mechanisms supporting the causal influence of SE on behavioural outcomes: (i) improved SE decreases anxiety and associated physiological arousal allowing the individual to

approach a task with less physical information; (ii) a person with high self-efficacy may be able to wilfully distract their attentions from potentially threatening physiological sensations (iii) the efficacious person may better able to persist, despite the perception and experience of pain and (iv) the efficacious person may be better able to re-interpret physiological sensations (Turk, 2002; Turk & Okifuji, 2002). Evidence suggests individuals who perceive they have control over their pain, who believe they can function despite pain and who avoid pain catastrophising function better than those who do not (Jensen & Karoly, 1991).

1.3.6 Social-cultural factors

The experience and expression of paediatric chronic pain is relationship and context specific. Individual pain beliefs and appraisals may be shaped by experience, modelling or information received from others (Vervoort, Goubert, Eccleston, Bijttebier & Crombez, 2006). Social factors such as social support and learning experience (e.g. parent modelling, peer acceptance) influence adjustment to chronic pain (Warrington & Younger, 2011; Vervoort, Huguet, Verhoeven & Goubert, 2011). In turn, learning experience may be integrated with individual trait characteristics which together predispose the individual with chronic pain to engage in adaptive or maladaptive coping. Walker and colleagues (2008) would suggest interpersonal relationships and mastery experience contribute to the individual's adoption of self-reliant, avoidant, dependent, infrequent or inconsistent coping approaches. In line with this, development theory suggests prior adaptation e.g. attachment might influence subsequent adjustment to chronic pain and thereby individual pain and coping profiles. Taken together, this research shows social support, modelling and learning experiences significantly influence function and coping behaviour over time (Chow, Otis, & Simons, 2016).

Parenting response to child pain has been examined through several constructs including protectiveness, minimisation and encouraging and monitoring responses (Claar, Walker, & Smith, 1999; Peterson & Palermo,

2004; Sieberg, Williams, & Simons, 2011; Palermo, Valrie, & Karlson, 2014; DuPen et al., 2016). Solicitous or protective parenting behaviours (e.g. exemptions from chores, attention and special privileges) have been shown to encourage illness behaviour (Langer, Romano, Levy, Walker, & Whitehead, 2009) and positively reinforce or exacerbate child pain (Noel et al., 2015; Evans et al., 2016). Child distress and behaviour and parent distress and behaviour can have an amplifying effect on each other with negative outcomes for the child and treatment response. For example, Logan, Simons and Carpino (2012) conducted mediational analyses to determine whether parental protective behaviour plays a mediating role between parental pain catastrophising and child school impairment. After controlling for known influences on child pain intensity and depression, parental pain catastrophising and parental protective response to child pain each independently predicted child school attendance rates and overall school impairment (Logan, Simons, & Carpino, 2012). Similarly, moderation analyses have shown parental response to child pain moderates child pain-catastrophizing and disability. Also, that the negative impact of catastrophizing on child disability was less pronounced when parents were highly engaged in the promotion of wellness coping responses (Vervoort, Huguet, Verhoeven & Goubert, 2011). Finally, parenting factors can have a longitudinal impact on child outcomes. Mediation and regression analyses have found parent distress (pain-related fear and pain catastrophising) was associated with child psychological and functional outcomes over time (Chow, Otis and Simons (2016). Parent protective behaviour was a significant predictor of child anxiety, depression, fear avoidance, disability and school functioning. Therefore, it is important parenting behaviour is considered in a treatment concurrent with child pain treatment.

Over-protective parenting may influence chronic pain in several ways. Firstly, over-protective behaviour may increase the child's perception of threat and reinforce a fear avoidant coping approach. Secondly, parental protectiveness may result in the child perceiving they have no control over their pain experience. Finally, over-protective parenting may exacerbate

chronic pain by limiting opportunities for accomplishment i.e. mastery of challenging situations may be prevented. Consequently, children may fail to develop a sense of self-efficacy for coping with chronic pain or to develop their repertoire of coping skills (Logan, Simons, Carpino, 2012). These effects are consistent with explanations of chronic pain identify fear of pain as the main determinant of the coping response (Asmundson, Noel, Petter & Parkerson, 2012).

1.3.7 The Fear-Avoidance Model

According to the fear avoidance model put forward by Lethem et al., (1983), avoidant or passive coping styles are associated with physical, social and emotional dysfunction whereas confrontational or active coping styles are associated with improved quality of life and functioning (Asmundson, Noel, Petter & Parkerson, 2012). The central premise of this model is that fear of pain resulting from movement or injury, leads to either a confrontational (adaptive) or avoidant (maladaptive) coping style. Vlaeyen et al., (1995) build on this by work by positing a cognitive behavioural model of fear avoidance. From this perspective pain perception and response is explained by a cycle of influence between affect (e.g. fear), behaviour (e.g. avoidance) and cognitions (e.g. beliefs). The authors emphasise the role of cognition as a core determinant of entering a negative pain cycle and ultimately shaping the pain response (see Figure 1.2). The fear avoidance model is supported by a large body of correlational evidence which supports these hypothesised links. However, this model is also criticized for a lack of clarity in explaining the direction of causality among variables or the role of pain-related anxiety in addition to the fear response (Leeuw et al., 2007). In addition, various types of anxiety e.g. physiological, cognitive, avoidance) are associated with varying degrees of physical disability and coping styles (McCracken & Gross, 1993). Despite these limitations the fear avoidance model was fundamental to the development of cognitive-behavioural treatment approaches to chronic pain (Turk & Okifuji, 2002; Gatchel, McGeary, McGeary & Lippe, 2014).

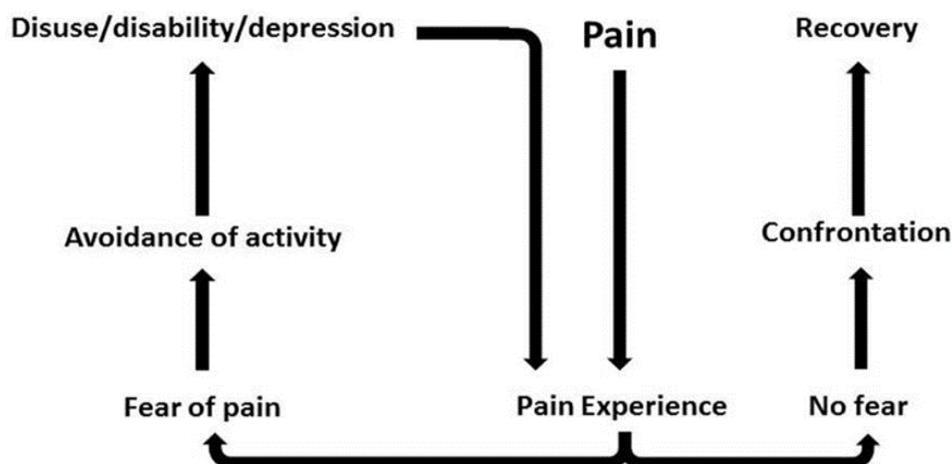


Figure 1.2 The Fear-avoidant Model of Chronic Pain (Vlaeyen & Linton, 2012)

In summary, chronic pain is a complex and multidimensional experience, influenced to varying degrees by a combination of factors including underlying pathophysiology and individual cognitive-affective, behavioural and sociocultural influences (Kerns et al., 2011). Each of these factors affect adjustment to pain and associated disability (Turk, 2002). The determinants of pain behaviour outlined above, have been incorporated in several theories of health behaviour change including Social Cognitive Theory (Bandura, 1996; 2005; 2006a). Social Cognitive Theory (SCT) is singled out as a potentially useful theoretical framework from which to understand chronic pain. SCT aligns with the biopsychosocial, tripartite model of influence outlined above and builds on more recent iterations of the FA model which place a greater emphasis on goals and self-regulatory processes to better explain individual differences in treatment response among chronic pain patients (Vlaeyen, Crombez, & Linton, 2009; Simons & Kaczynski, 2012; Fisher & Palermo, 2016). It is from this conceptualization that the current theoretical understanding of chronic pain is deduced.

1.3.8 Social Cognitive Theory

Bandura's (1996; 2006a) Social Cognitive Theory (SCT) is an extension of his earlier social learning theory (Bandura, 1971; 1977a). In addition to behavioural principles and developmental perspectives, SCT also incorporates an understanding of cognitive influences, observational learning and social context on human behaviour. SCT posits that individual behaviour may be understood in terms of human capabilities including the ability to engage in forethought, self-regulatory and self-reflective practice, symbolic thinking and vicarious learning processes (Bandura, 1996). Bandura (1996) suggests these capabilities are nurtured or neglected depending on social practice rather than chronological age. This implies a process of reciprocal causation between interacting determinants of human behaviour.

Bandura (1989a; 1996) describes this causal process of behaviour change as a network of reciprocally interacting domains of influence, namely person, behaviour and environment (see Figure 1.3). These domains of influence, each represent separate and interlinking determinants of behaviour that work in tandem to influence behaviour change (i.e. reciprocal causation) (Bandura, 2005; 2006a). These separate sets of psychological processes are experienced by individuals to varying degrees and affect different outcomes (Bandura, 2005). Person based processes of behaviour change include cognitions, affect and biologic events. These determinants of behaviour are further explained in terms of individual beliefs, self-efficacy and outcome expectations (Bandura, 1997b). Behaviour based processes of behaviour change refer to individual habits and behaviours including self-regulation, behavioural capability, behavioural goals (Bandura, 1989b; 1991). Environmental processes of behaviour change include social practice or context and individual experience or perception of social response, support, facilitation and observational learning (Bandura, 1996; 2005; 2006a). Understanding these separate components and how to target them may be important for the development of an effective treatment intervention.

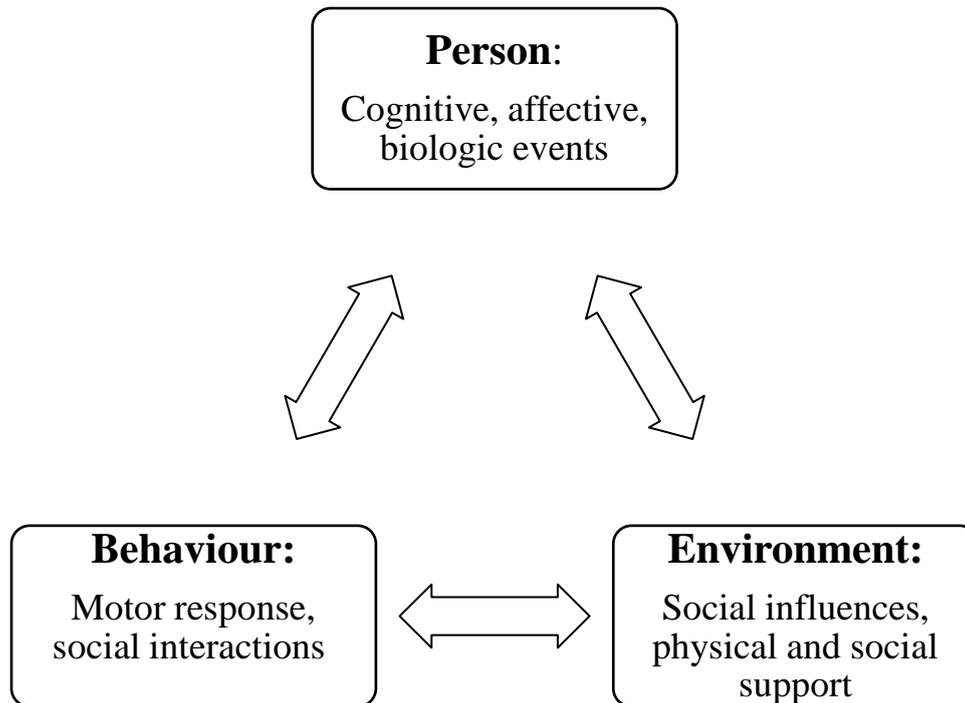


Figure 1.3 Conceptualisation of reciprocal determinism in Social Cognitive Theory (Bandura, 1996).

1.3.9 How does this apply to coping behaviour?

Reciprocal causation between person-behavioural factors refers to the interaction between thought, affect and action. As to chronic pain, an individual's cognitions (negative outcome expectations, self-efficacy beliefs) shape and guide their response to pain. In turn, individual actions (fear-avoidance, passive coping) bi-directionally influence thought patterns (e.g. pain catastrophising) and emotional reactions (e.g. low mood). Person factors including biological properties (e.g. physical structure, sensory and neural systems) and developmental competencies (e.g. self-regulation, forethought) impose constraints on learning capability and behaviour acquisition (Bandura, 2006b). These systems are in turn modifiable by

behavioural experience (e.g. observational learning, social influence) (Greenough, Black & Wallace, 1987).

Reciprocal causation between behaviour-environment factors may be described as the dealings of everyday life whereby an individual's behaviour alters their environment and is in turn altered by the conditions it creates. Most aspects of the environment only exert influence when they are activated by behaviour. In the context of chronic pain, for example, over-protective parental behaviour may be elicited by a child's expressions of pain and vice versa (Lynch-Jordan, Kashikar-Zuck, Szabova, & Goldschneider, 2013). Furthermore, the child with chronic pain might transform their environment through their behaviour via the selection and creation of activities, situations or associations based on their preferences and competencies (Bandura, 1989a; 1996).

Reciprocal causation between environment-person factors refers to the modifying effect of social influences on outcome expectations, beliefs, emotional predispositions and cognitive competencies. Social influences including social reaction convey information and trigger an emotional response. This may be achieved through modelling, instruction and social persuasion (Bandura, 1989). Social reaction can vary depending on physical characteristics (e.g. age, sex, race) and social role or status (e.g. assertive, aggressive). In turn, social reactions (e.g. being treated differently) affect the individual's conception of themselves (e.g. self-efficacy) and others in a way that strengthens or alters the individuals experience of their environment (Greenough et al., 1987).

Is this developmentally appropriate?

SCT offers a developmental perspective on the process of learning and behaviour acquisition across the lifespan which might be applied to a younger chronic pain population (Bandura, 2006b). According to SCT, cognitions are a mediator of individual behaviour and of learning processes which include attention, retention, reproduction and motivation. These

processes are important considerations for learning and behaviour acquisition in early development and across the lifespan. Thus, this framework may be particularly suited to guide the development of an intervention for a pain population where cognitive competency is still developing, and social influences are considerable.

Interventions are being developed for older children and adolescents and the needs of younger children are being obscured (Fisher et al., 2014; Fisher, Law, Palermo & Eccleston, 2015). This is a gap in the literature which may be addressed by incorporating developmental, learning-focused perspectives in intervention development. SCT has successfully been used to develop interventions on dietary behaviour among children (Rolling & Young Hong, 2016) and to promote physical activity among women (Jang & Yoo, 2012; Stacey, James, Chapman, & Lubans, 2016; Joseph, Ainsworth, Mathis, Hooker, & Keller, 2017). However, a review of the literature suggests SCT has never been applied as a guiding theoretical model for an intervention targeting school-age children with chronic pain.

A theoretical understanding of the mechanisms of behaviour change should inform the selection of intervention components most likely to address the determinants of behaviour and engage the target population. For example, an intervention targeting knowledge and coping skills (e.g. behavioural capability) among children with chronic pain would use pain education and supported skills training (e.g. modelling, guided discovery, positive reinforcement, thought-challenging) to promote self-regulation, problem solving. Chapter 2, 4 and 5 illustrate this process in greater detail. Testing this framework may prove useful in highlighting and explaining pathways to adaptive coping behaviour change in this population.

1.4 Section III: Management of chronic pain

The focus of this thesis is the adaptation and feasibility evaluation of an online version of the *Feeling Better* programme. Several issues for consideration are noted from the outset.

1.4.1 Paediatric chronic pain: assessment and outcomes

It is unknown if Internet-mediated assessment and treatment of pain and associated characteristics are acceptable to and feasible for delivery to younger children. Specifically, it is unknown if the limited range of validated psychometric tools relevant to this research are a good fit for remote methods of assessment and evaluation of response to therapy. The PedIMMPACT committee recommend core outcomes e.g. physical functioning and a range of associated psychometric measures that should be used in clinical trials to promote clarity and comparison of findings (McGrath et al., 2008). The recommended measures are robust but generic instruments that may be supplemented with specific measures consistent with the theoretical basis and model of therapy provided e.g. the pain catastrophising scale (Crombez et al., 2003). Types of psychometric measures have been identified as appropriate according to age and development level. For example, facial expression scales such as the Faces Pain Scale Revised (Hicks, von Baeyer, Spafford, van Korlaar, & Goodenough, 2001) are appropriate for younger children whereas visual analogue and numerical scales can be used effectively by middle school age children (Chambers, Giesbrecht, Craig, Bennett, & Huntsman, 1999; Von Baeyer, Uman, Chambers, & Gouthro, 2011; Palermo et al., 2012). Pain assessment research has shown children can appreciate the influence of pain and rate pain intensity at five years of age and at 7 years of age children can describe the quality of their pain (Cohen et al., 2008; McGrath et al., 2008; von Baeyer, Lin, Seidman, Tsao, & Zeltzer, 2011; von Baeyer et al., 2011; Palermo et al., 2012).

1.4.2 Evidence based treatment for chronic pain

The “gold standard” treatment for chronic pain is a multidisciplinary approach involving pharmacological and non-pharmacological disciplines with a focus on improving function across all domains (e.g. physical, emotional, social and role functioning) (Turk et al., 2011). Comparison of

conventional medical treatments and comprehensive pain programmes have found the latter to be the more efficacious, cost-effective and evidence-based approach (Gatchel & Okifuji, 2006). For individuals with pain related disability or associated co-morbidities an interdisciplinary treatment approach is optimal. This can include psychological therapy, physical and occupational rehabilitation and standard medical care. Psychological aspects of the pain experience indicate a need for psychological intervention as part of a multidisciplinary or interdisciplinary treatment approach (Kerns, Sellinger, & Goodin, 2011; Morley & Williams, 2015). Psychological treatment may include cognitive behavioural therapy, biofeedback, relaxation training, mindfulness, acceptance and commitment therapy, psychotherapy or a combination of these approaches. The evidence base supporting the use of cognitive behaviour therapy for chronic pain management is arguably the most robust (Morley, Williams, & Eccleston, 2013).

1.4.3 Cognitive behaviour therapy for chronic pain

The efficacy of cognitive behavioural therapy has been extensively evaluated in empirical treatment studies involving adults and children, in traditional face to face settings and using remote delivery methods. This evidence base has been described as underwhelming despite the wealth of research focused on this issue (Eccleston & Crombez, 2017). There are several systematic reviews of treatment effectiveness that identify CBT as a promising therapeutic approach for adults and children with chronic pain. The highest quality of which are the four main Cochrane Systematic Reviews of psychological therapies for adults and children with chronic pain conducted in traditional face to face and Internet-delivered treatment settings (Eccleston, et al., 2014a; Eccleston, et al., 2014b; Fisher et al., 2015). This evidence base identifies behavioural and cognitive-behavioural treatments as moderately effective for adults with chronic pain in comparison with waiting lists on measures of pain, disability and mood (i.e. absolute efficacy). However, in comparison with active control conditions,

psychological therapies are associated with small or no effects on these outcomes (i.e. relative efficacy). The relative efficacy of CBT is dependent upon the measures used and the nature of the trial. Small to moderate effects sizes for CBT as based on an amalgamation of studies with high heterogeneity of key study parameters (e.g. sample, control arms, diagnosis and outcome measures), therapeutic content, treatment exposure and operator characteristics (Morley, 2011; Eccleston, Morley, & Williams, 2013; Eccleston & Crombez, 2017).

Evidence from systematic reviews and meta-analyses suggest the efficacy of psychological therapies for child and adolescent chronic pain is of low to moderate-quality. Efficacy data provide preliminary support for the use of CBT in support paediatric pain management, specifically chronic headache and increasingly musculoskeletal pain conditions (e.g. fibromyalgia) (Eccleston et al., 2013; Morley et al., 2013). However, these findings should be interpreted with caution. Much of this research is of exploratory and based on small sample sizes. In comparison with CBT for adults, evaluations of efficacy for paediatric pain populations are less conclusive.

In a pilot and subsequent full-scale randomised controlled trial of CBT for paediatric chronic pain, Palermo and colleagues (2009; 2016) found Internet-based CBT was associated with improved outcomes across pain conditions including headache, abdominal pain and fibromyalgia (Palermo, Wilson, Peters, Lewandowski, & Somhegyi, 2009; Palermo et al., 2016). It is associated with significant improvement in pain coping strategies and reduction in pain catastrophising in adolescents with juvenile fibromyalgia (Kashikar-Zuck, et al., 2013b). Clinical trials have found CBT to be significantly more effective than pain education in reducing functional disability in adolescents with juvenile arthritis and fibromyalgia (Kashikar-Zuck et al., 2012a; Kashikar-Zuck, et al., 2013b). In contrast, several pilot RCT's of remotely delivered CBT for pain management in adolescents with chronic pain reveal no significant differences between treatment and waitlist control groups on measures of paediatric quality of life among children with

recurrent abdominal pain or headache (Hicks, von Baeyer, & McGrath, 2006) or self-reported depression and parental protectiveness among adolescents with mixed chronic pain conditions (Palermo et al., 2009). Comparison of the CBT interventions is difficult because CBT was seemingly applied very differently, and the intervention components associated with estimates of effect were not identified.

The scope of CBT is broad and encompassing. The contemporary CBT model has been influenced by classical and operant conditioning (Fordyce et al., 1973), the stress- appraisal response model (Lazarus, 1966; Turk, 2002), cognitive learning theory and therapy (Beck & Weishaar, 1989; Clark, Beck, & Alford, 1999), mindfulness (Kabat-Zinn, Lipworth, & Burney, 1985; Kabat-Zinn, 2006), social learning theory (Bandura, 1977a) and the fear avoidance model (Vlaeyen et al., 2009; Crombez, Eccleston, Van Damme, Vlaeyen, & Karoly, 2012). Current applications of CBT for chronic pain management may be comprised of some variation of the following therapeutic components: pain education, goal setting, relaxation training, graded activity, operant principles, attention management, problem-solving, cognitive restructuring, mindfulness, behavioural experiments and future planning (Vlaeyen & Morley, 2005; Morley & Williams, 2015). These components combine to create a problem-focused, goal-oriented approach to coping.

As stated previously, patient beliefs and appraisals are considered the primary determinant of the behavioural and psychological response to pain however causality remains unclear (Yu et al., 2012; Morley et al., 2013; Levy et al., 2014). According to the cognitive behavioural model, the mechanisms of change are separate and interacting changes in individual cognition, emotion and behaviour. CBT strategies are designed to help individuals systematically address maladaptive cognitions, emotions and behaviours (Turner, Mancl, & Aaron, 2006). Further insight into the therapeutic mechanisms supporting patient improvement in response to CBT might be achieved through higher standard of trial reporting. Explicit report of intervention development and application of theory may facilitate

comparison of intervention components associated with beneficial treatment effects. More research is needed that systematically and transparently applies an identified CBT model; that accounts for the influence of parenting in the treatment of paediatric chronic pain; one that explores treatment response in terms of individual differences and specific populations; that embraces the potential of technology as a support for traditional treatment and the delivery of novel therapeutic content (Eccleston & Crombez, 2017).

1.4.4 Coping response

Coping has been described as the cognitive and behavioural effort to manage external and/or internal demands that are appraised as stressful and exceeding the resources of the individual (Lazarus & Folkman, 1984; Schneiderman, Ironson & Siegel, 2005). Cross-sectional and longitudinal studies demonstrate adaptive coping is associated with short term and long-term adjustment to chronic pain (Jensen et al., 1991; Turner et al., 2006; Kashikar-Zuck, et al., 2013b). Maladaptive coping is negatively associated with adjustment. Some regression analyses suggest that maladaptive coping responses accounted for more variance in adjustment to chronic pain than the adaptive responses (Tan, Teo, Anderson, & Jensen, 2011). These findings suggest children with chronic pain would benefit more from stopping or reducing maladaptive coping responses than from increasing the use of adaptive coping responses (Tan & Jensen, 2008). Geisser, Robinson and Riley (1999) suggest the likelihood of engaging in pain coping strategies is influenced by feelings of helplessness and perceptions of control over pain. It follows that if maladaptive coping is reduced and adaptive coping behaviour is established in childhood, the long term impact of chronic pain may be diminished (Geisser et al., 1999). Maladaptive coping can include pain catastrophizing, fear-avoidant, passive or dependent coping behaviour. Pain-related beliefs, appraisals and coping are the basis of cognitive behavioural approaches to pain management.

1.4.5 CBT for school age children

Questions remain regarding the use of CBT for children, particularly for early and middle school age children. Early school-aged children (5–7 years) and middle school age children (7-12) are particularly under-represented in CBT outcome literature (Grave & Blissett, 2004). Response to treatment in this sub-group is important to understand because age and developmental level may moderate treatment outcomes. Evidence for the efficacy of CBT for pre-adolescent children with chronic pain is inconsistent and unclear. Treatment studies in this field have typically focused on adolescents with chronic pain or involve small samples with broad age ranges spanning pre to late adolescence. Only one treatment study was found that both limited recruitment to a pre-adolescent age range and evaluated the efficacy of a CBT programme (Lelieveld et al., 2011). CBT was used to promote physical activity in children ages 8-12 years with juvenile idiopathic arthritis. The programme was delivered using combined in-person and online methods. Absolute efficacy was evaluated and improvements in PA were observed in both arms, however only the CBT intervention was associated with improvement among those with low PA at baseline. Importantly for the age group, the intervention was also found to be safe, feasible and had good adherence.

Research exploring the role of child age in relation to treatment outcomes is sparse and largely focused on childhood anxiety disorders (Grave & Blissett, 2004; Freeman et al., 2007; O'Connor & Creswell, 2008). This literature presents mixed findings regarding the importance of child age. Some treatment studies for child anxiety disorders suggest age has no effect on outcomes whereas others suggest younger children do less well in CBT compared to older children and adolescents. Much of this research is focused on participants between 8-12 years of age (Freeman et al., 2007). One exception is a non-controlled study of CBT for 3-7-year-old anxious children. Significant improvements were observed in symptom severity and functioning from pre to post-treatment following an average of 8.3 treatment sessions (Minde, Roy, Bezonsky, & Hashemi, 2010).

To date, no CBT-based chronic pain studies have included or focused on children with chronic pain who are younger than 7 years of age. This may be because middle childhood (7- 12 years) is associated with the development of deductive reasoning abilities, and rapid development in logic and perspective taking skills (Piaget, 1964; Piaget & Inhelder, 1969; Ojose, 2008). At this stage children begin to understand that others may have different thoughts to their own (i.e. decreased egocentrism). In addition, children aged 7-12 years begin to develop problem solving and meta-cognitive skills that may allow for a better treatment response (Kingery et al., 2006). The sensitivity of CBT for younger children is an important issue to address given the prevalence and trajectory of paediatric chronic pain (Mulvaney et al., 2006; King et al., 2011). In addition, the findings from CBT-based treatment studies comprised mostly of adolescents may not generalise to younger children due to differing capabilities (Nelson & Tusaie, 2011). Differences in symptom expression, self-report and treatment response may be obscured in studies involving a wide age range. CBT adapted to pre-adolescent children with chronic pain may help younger children understand the relationship between their thinking, behaviour and experience of chronic pain and to find the words to express their understanding of this relationship to others (Friedrichsdorf et al., 2016). Moreover, differences in the symptom profile of children and adolescents with chronic pain could have important implications for treatment response and tailoring.

Research suggests the optimal application of CBT for younger children is one where developmental theory are integrated with cognitive behavioural theory and creative methodologies, such as the use of narrative and analogy (Grave & Blissett, 2004). The specific CBT strategies used for children with chronic conditions are derived from those used in the treatment of adults. CBT strategies may be adapted to the developmental level of the child with a greater focus on behavioural strategies to accommodate cognitive capabilities. Some cognitive elements of CBT can be complex and require more abstract thinking. For younger children

behavioural strategies may be more useful as they offer a way to target a problem through more concrete information processing. CBT treatment may begin with psychoeducation to inform children about the nature of chronic pain. Relaxation strategies may be introduced to address associated physiologic and somatic complaints. Cognitive strategies such as positive self-talk, coping statements and cognitive re-structuring may then be used to help children identify and challenge maladaptive thinking related to the experience of pain. Behavioural strategies might include graded exposure and problem-solving to change avoidant responses to situations associated with the maintenance of pain symptoms. Finally relapse prevention strategies may be presented to help the child cope with future pain flare ups or challenging situations. Overall CBT aims to assist the child manage their pain, improve function despite pain and overall quality of life.

1.4.6 Early, self-management support

Given the long-term nature of chronic pain conditions, psychological interventions identify pain self-management as integral to treatment success. There are many definitions of self-management (SM), here it is described as the process through which the individual learns to incorporate the skills and knowledge to provide self-care and become an active participant in the care process (Barlow et al. 2002; Lorig & Holman, 2003, Whittmore et al., 2008). In the context of chronic pain, self-management begins with an acceptance of a coping rather than curing treatment approach. Successful pain self-management must then involve knowledge and coping skill acquisition, improved self-efficacy for task performance and enhanced sense of personal agency (Barlow, Wright, & Sheasby, 2002). Studies exploring the effectiveness of self-management as a treatment approach for chronic conditions are difficult to evaluate due to inconsistency in study parameters (Carnes et al., 2012). Evidence to support the short-term effectiveness of self-management interventions is promising, however the long terms effects of self-management behaviour have not been sufficiently researched. There are also gaps in our understanding of the

most effective, cost-effective and sustainable course components (Du et al., 2011; Carnes et al., 2012; Sell, Amella, Mueller, Andrews, & Wachs, 2016). Despite these limitations, epidemiological data consistently confirm children with chronic pain have poorer outcomes on functioning and quality of life measures compared to the general population. Thus, effective self-management interventions are indicated (Palermo et al., 2012).

Due to the complex nature of chronic pain and to prevent maladaptive coping strategies becoming entrenched, early management of pain symptoms and associated disability is also recommended (Palermo et al., 2012). However, individuals with chronic pain often do not benefit from access to psychosocial interventions that might facilitate pain self-management. Psychological support for chronic pain management is neither routinely offered nor readily available (Lynch et al., 2007; Lynch, Campbell, Clark et al., 2008; Palermo et al., 2019). It is common for pain self-management to be promoted as a treatment approach even before diagnosis of the chronic pain condition or access to pain self-management training. Treatment might begin with pharmacologic intervention initiated at the primary care level and referral to a pain specialist. For adults and children with chronic pain, reasons for poor pain self-management and uptake of psychological support include lack of access to trained healthcare providers or long waiting lists (Peng et al., 2007; Lynch et al., 2007; 2008; Guerriere et al., 2010).

Barriers to engagement and use of psychological services also include lack of knowledge about this treatment approach (Darnall et al., 2016), stigma associated with the use of psychological support (De Ruddere & Craig, 2016; Williams, 2016), geographical or financial barriers, and conflicting family or work commitments (Jerant, Von Friederichs-Fitzwater, & Moore, 2005; Peng et al., 2007). For example, patients living in rural areas in the Republic of Ireland may have to travel two to three hours each way to attend secondary care, pain clinic appointments and the waiting list to access psychological support from a trained therapist may be up to two years (CRA, 2018). Attendance is further complicated by full-time

employment, school or family commitments and difficulty travelling due to disability. The effect of waiting for treatment for six months or longer include deterioration in overall quality of life, psychological well-being and an increase in depression scores (Lynch et al., 2007; Palermo et al., 2019). For younger children these issues may be compounded by developmental barriers in terms of cognitive or emotional capacity; perceived lack of control; non-disclosure of pain or symptoms; lack of knowledge, maladaptive pain beliefs and characteristics or patterns of behaviour (McGrath & Frager, 1996). This has led to the development of Internet-mediated psychosocial treatment for a range of patient populations. It may be that web-applications are a solution to logistical and other barriers that limit access to pain management information and training.

1.4.7 Internet-based psychological interventions for chronic pain

The 2012 American Pain Society Position Statement, “Assessment and Management of Children” identified the development and evaluation of novel treatment strategies and innovative delivery methods as a key area for future research development (Palermo et al., 2012). The field of Internet-mediated intervention development for paediatric chronic pain is relatively new and dynamic. Despite this, there are a multitude of definitions for internet mediated platforms which facilitate the delivery of healthcare information and training. The terms used to describe these platforms are used interchangeably across the literature e.g. eHealth, information and communication technology (ICT), mobile or m-Health, digital health interventions (DHIs) and telehealth. The WHO defines eHealth as “the cost-effective and secure use of information communication technologies (ICT) in support of health and health-related fields, including health-care services, health surveillance, health literature, and health education, knowledge and research”. Some argue that like the Internet, eHealth should be defined in terms of how it is used (Eysenbach, 2001). The current study uses the term information and communication technology (ICT) which includes all communication technologies regardless of application, device or system.

Internet-based psychological therapies supporting pain management for children and adults with chronic pain have demonstrated promising results, but efficacy data is inconclusive (Eccleston et al., 2014a; Fisher et al., 2015). In recent years, a Cochrane Systematic Review found the remotely delivered psychological therapies for young people with chronic pain may be efficacious, particularly for children with chronic headache (Fisher et al., 2015). However, research in this area is in its infancy, there are few treatment studies, that evaluate the effectiveness of online interventions for paediatric pain management and most are exploratory in nature. Participants in the Internet intervention groups have shown significantly greater reduction in activity limitations (Connelly, Rapoff, Thompson, & Connelly, 2006; Palermo et al., 2009; Palermo et al., 2016) and clinically significant improvement at follow up compared to active and waitlist control group peers (Connelly et al., 2006). However, clinical outcomes such as pain intensity, mood and quality of life have been found to be more resistant to treatment (Hicks et al., 2006; Palermo et al., 2009; Trautmann & Kröner-Herwig, 2010). ICT-based, psychosocial interventions have been developed and evaluated for adolescents with mixed chronic pain (Palermo et al., 2009; Palermo et al., 2016); recurrent headache and migraine (Connelly et al., 2006; Trautmann & Kröner-Herwig, 2008, 2010; Rapoff et al., 2014) and juvenile arthritis (Stinson et al., 2010a). These treatments are typically brief, 4-12 weeks in duration, based cognitive behavioural therapy and feature minimal contact with an online therapist or tutor. Recent Cochrane systematic reviews show these studies tend to include a wide age range or focus exclusively on adolescents. Also, the involvement of parents in remotely delivered therapy has been limited. These interventions are discussed in greater detail in Chapter 3.

If feasible and effective, ICT-based therapy may be used to reduce attendance at pain clinics or as an adjunct to treatment as usual. It may be used to support patients currently on the waiting list or to provide booster sessions for those who have completed their allocated course of therapy. This is an important advantage. In the Republic of Ireland for example, the

recently published, Children's Rights Alliance Report Card revealed demand for mental healthcare is exceeding availability at all levels (childrensright.ie, 2018). The report states that at a primary care level, there are 6,811 children under the age of 17 are waiting to access community-based psychology services by the end of July 2017. Four out of five of these children are under the age of fifteen and a third had been waiting over one year. The demand for mental health services has been increasing with referrals in 2017 11.3% higher than 2016. The specialised Children and Adolescent Mental Health Services (CAMHS) report an 8% increase in children waiting for a first appointment. Given the pressure on services like CAMHS, the UN Committee have recommended that out of hours services for children be prioritised and strengthened in a revision of mental health policy (United Nations, 1989; Children's Rights Alliance, 2018).

Advantages of ICT-delivered interventions include reduced costs and increased access and convenience for users. Interventions delivered using the Internet (eHealth) and mobile devices (mHealth) have the advantage of offering "real-time" momentary data collection and self-management support. Internet-based therapy delivery may address the logistical and financial demands preventing access to treatment. Remotely delivered treatment has the advantage of empowering the individual users in pain self-management when most needed and at the patient's convenience. Moreover, pain management training in the home may foster the habit of regular practice and normalize the process of active coping for the individual and family. In addition, convenient access to evidence-based psychological support for paediatric pain management may overcome social and psychological barriers such as lack of knowledge and perceived stigma associated with accessing psychological treatment.

The disadvantages of remote treatment delivery can include the absence of direct contact with a therapist, the need for self-direction and its reliance on physical access to the internet. ICT-based therapy is associated with reduced opportunity for spontaneous clarification by the therapist if the child is having difficulty completing a strategy (Rochlen, Zack, & Speyer,

2004). Guided self-management programmes can involve remote access to a tutor or coach during the active phase of the programme. This is no replacement for the therapist client relationship. The literature is mixed as to whether “supported” or “unsupported” self-management interventions are more effective. Recent studies suggest supported ICT-based interventions are associated with slightly larger effect sizes compared to unsupported (Carnes et al., 2012; Morrison, Yardley, Powell, & Michie, 2012; Morrison, Moss-Morris, Michie, & Yardley, 2014a; Morrison, 2015; Yardley et al., 2016). The autonomy afforded by remote delivery may have the adverse effect of making child on a waiting list feel side-lined. Unlike face to face therapy, online treatments offer no opportunity for learning from visual or social cues that may be derived from therapists modelling of CBT strategies. One way to address this issue is to include video recordings of demonstrations of behaviour and periodic feedback on progress. Finally, the nature of online interventions is such that regular and reliable access to the Internet is a necessity. This means those without access to the Internet or a reliable Internet connection are at a disadvantage. More research is needed to evaluate the feasibility and acceptability of Internet-mediated therapy delivery in different contexts.

1.4.8 Feeling Better

Feeling Better is a cognitive behaviour therapy-based pain management manual which was developed for carers of individuals with chronic pain and intellectual disabilities (McManus & McGuire, 2010). The programme is designed to promote pain self-management using evidence-based cognitive-behavioural strategies. The manualised pain management programme was previously evaluated in a pilot trial and group-based setting for people with chronic pain and intellectual disabilities (Kennedy, O’Higgins, Sarma, Willig & McGuire, 2014; McManus, Treacy & McGuire, 2014; Kennedy, 2016). Preliminary evidence of the effectiveness of the programme is provided by authors. The utility of the programme was tested in a case series evaluation involving five individuals with chronic

pain and mild range intellectual disability. Participants received 8 sessions of CBT based on components of the programme. Use of the manualised programme was associated with improvement in pain management knowledge, well-focused coping and coping effectiveness at post-treatment but not at follow-up (McManus, Treacy & McGuire, 2013). The potential effectiveness of the programme was further tested in a mixed method, matched controlled clinical trial of the *Feeling Better* programme designed to support young women with dysmenorrhea and intellectual disabilities (Kennedy, O'Higgins, Sarma, Willig & McGuire, 2014; Kennedy, 2016). This trial evaluated the effectiveness of the programme for menstrual pain management in young women (N = 32), aged 12 – 30 years (M= 18, SD = 4.89) who have a mild - moderate intellectual. Participants were assigned to a 12-week Feeling Better pain management group (n = 18) or a treatment as usual control condition. Participation in the pain management group was associated with a significant increase in pain management knowledge over time ($p = .003$) compared to the control group at post-intervention but not at follow-up. No statistically significant effect of group or time was observed however, participation in the pain management programme was associated with small improvements in pain intensity, pain interference, use of pain coping strategies and parental ratings of these constructs. The promise of these findings was considered sufficient to warrant further research. The *Feeling Better* pain management programme has never been evaluated in an online setting.

In comparison with other manualised pain management programmes (e.g. Palermo, 2012; Murphy, et al., 2014) the *Feeling Better* programme may be particularly suited to a paediatric pain population and to novel technology-mediated delivery methods for several reasons.

First, the *Feeling Better* programme may be suited to the target pain population because it is more focused on behavioural strategies in line with the developmental capabilities and needs of the intended patient group. Moreover, the programme was written to be delivered by care-givers for individuals with chronic pain and intellectual disabilities. The manualised clinical content is intended to be used by care-givers with little to no pain

management training for individuals with low literacy levels and for whom ease of understanding is a priority. Therefore, the text-based content and visual illustrations are informal in presentation and may be more easily adapted to the appropriate reading and intervention intensity levels.

Second, there are no other treatment studies or manuals that evaluate the effectiveness of psychological therapy, delivered remotely and dedicated to pre-adolescent children with chronic pain. Increasing this knowledge base may be important to meet the needs of these patients. Given the novelty of the delivery method, the clinical content should derive from psychological therapy supported by the best-available evidence. As stated previously, cognitive behaviour therapy is currently the most researched and robust treatment approach.

Third, the *Feeling Better* programme is a modularised programme designed to support pain management from home. As such it may be suited to web-based delivery and website platforms without compromising the integrity of the treatment.

Finally, the manualised *Feeling Better* programme encourages caregivers to act as lay-therapists for the individual with chronic pain. Given the links between parenting behaviour and the management of chronic pain in children, it seems logical that parents should be involved in the treatment approach. Involving parents in the treatment approach is more likely to increase their commitment to it and encourage the parent to act as a facilitator of the therapy (Stallard, 2009; Richardson, Stallard, & Velleman, 2010). Parents may be taught strategies to assist their child in the management their pain and educated as to how their behaviour can influence child pain and coping behaviour.

1.4.9 Conclusion

This summary of the literature has identified several gaps between evidence-based recommendations and current practice in supporting paediatric pain self-management. Specifically, this literature review identified the need for research to focus on the development and evaluation of effective, evidence-based, developmentally appropriate and widely

accessible interventions for paediatric chronic pain management. The current adaptation and evaluation of the *Feeling Better* pain management programme may provide valuable information to address these gaps in the literature.

1.4.10 Overall aim

The overall aim of this research was to create a developmentally appropriate and widely accessible version of an existing, CBT-based pain management programme called *Feeling Better* (McManus & McGuire, 2010). This involved the adaptation of the original, manualized CBT programme for an online mode of delivery and for a much younger pain population.

1.4.11 Research questions and thesis outline

This research was designed to test the hypothesis that an evidence-based, theory-informed approach could be used to adapt an existing manualised and adult-focused CBT intervention to be acceptable to pre-adolescent children with chronic pain and merit progression to a full scale randomised controlled trial (RCT) of effectiveness. To investigate this hypothesis, four research questions were generated:

1. What is the effectiveness of psychological therapies for paediatric chronic pain management delivered using information and communication technology?
2. What are the experiences, barriers and facilitators of pain self-management from the perspective of the pre-adolescent children with chronic pain and their care-givers?
3. How do younger children with chronic pain (5-12 years) and their care-givers perceive ICT-based therapy delivery and what are the design features and intervention components most likely to engage this population?
4. Is it feasible to develop and perform a randomised controlled trial of

an Internet-based version of the *Feeling Better* programme and how would such a programme be used by families coping with chronic pain?

Each of these research questions were investigated in separate but inter-linking studies. The result of which is an overall body of research which is multi-phased and mixed method (see Table 1.1). A parallel rather than linear approach was taken in completion of these studies. This process is outlined below and described in detail in Chapter 2.

1.4.12 Ethical Considerations

Each of the studies contributing to this body of research were approved by the National University of Ireland Galway Research Ethics Committee (NUI Galway Ref no. 13/Nov/01) (see Appendix 1). Participants in each study were given a Participant Information Sheet and provided informed consent and assent. All participants were made aware that their participation would remain confidential and that they were free to withdraw from the study at any point without providing a reason for doing so. All participants were de-briefed at the close of their participation in each study and provided with de-briefing forms. Due to the vulnerable nature of the participants, special care was taken to follow the recommended code of conduct regarding informed consent and assent. Contact information for the researcher and further sources of support, information and counselling were made available to all participants.

1.4.13 Thesis format

This thesis takes the form of 7 chapters summarised below and outlined in Table 1.1.

Chapter one

Chapter one provides a broad overview of the literature relevant to paediatric chronic pain, assessment, treatment and management, cognitive behavioural therapy and appropriate theoretical frameworks. This includes a summary of current gaps in knowledge relating to the mechanisms of chronic pain, novel treatment strategies and innovative delivery methods. This chapter closes with an overview of the format of the thesis.

Chapter two

Chapter two of this thesis focuses on the methodology and development of the online intervention for this study. It outlines the application of “best practice” guidelines and an intervention mapping protocol for intervention development. This includes the application of Social Cognitive Theory (Bandura, 1996; 2005) and a model for the development of Internet interventions (Ritterband, Thorndike, Cox, Kovatchev & Gonder-Frederick, 2009) as a guiding framework for the selection of intervention components. This phase of research functions as a guide subsequent phases of intervention development and evaluation.

Chapter three

Chapter three of this thesis is a systematic review, meta-analysis and intervention content analysis of the pain literature. The aim of this research was to answer research question 1. This evaluation focuses on the effectiveness of ICT-based psychological treatment for paediatric chronic pain and incorporates recent publications. The role of ICT-based psychological treatment for paediatric pain management is discussed in terms of treatment effectiveness and intervention development and report.

Chapter four

Chapter four presents the results of the qualitative needs assessment conducted to understand pain self-management from the perspective of pre-adolescent children and their care-givers. This was a qualitative study using participative research methods and theoretical analysis. The study incorporates potential end-user input, evidence from the literature and relevant theory to inform current understanding of the contextual factors that shape the lived experience of chronic pain management. These experiences are explored from the perspective of pre-adolescent children and their care-givers. This study employs a theoretical analysis of qualitative data that informed the development of the proposed pain management intervention. The aim of this research was to answer research question 2. The adaptation of the *Feeling Better* pain management programme began in 2014 and was developed and tested between 2015 and 2017.

Chapter five

This chapter outlined the focuses on the development and usability testing of the adapted, online version of Feeling Better pain management programme prior to the feasibility trial. This research is designed to answer research question 3. Key design features and intervention components that appeal to younger children with chronic pain and their caregivers are identified and the results of the development process are evaluated in usability testing, incorporating real end users' input, evidence from the literature and the experience of an expert panel.

Chapter six

Chapter six presents the research results of a two-armed feasibility trial and the final study in this body of work. Feasibility is evaluated in terms of recruitment, adherence, attrition and treatment satisfaction. Exploratory analyses of absolute efficacy are also presented. The effect of the guided

Internet intervention on overall quality of life (physical and psychosocial), pain intensity, self-efficacy, pain catastrophising and parental protective behaviour compared to a waitlist control are evaluated. The aim of this research is to answer research question 4.

Chapter seven

The final chapter of this thesis is an interpretation of the overall findings. Trial strengths and limitations are discussed, along with validity and generalizability of the study. This research is compared to similar interventions and those published since the inception of this study. Finally, this thesis work is summarized, noting implications for practice and future research.

Table 1.1

Overview of aspects studied, aims and development process

	Study 1 (Chapter 3)	Study 2 (Chapter 4)	Study 3 (Chapter 5)	Study 4 (Chapter 6)
Aim	Evaluate the effectiveness of remotely delivered interventions for paediatric chronic pain	Explore child and parent experiences of pain self-management	Explore user preferences and perceptions of digital health interventions	Explore feasibility and efficacy of Internet-based psychological intervention compared with usual care Waitlist control on feasibility and clinical outcomes
eHealth intervention	Hardcopy draft	Standalone web- pages and PowerPoint graphics	Feeling Better 1.0, website prototype	Feeling Better 2.0 website
Method	Systematic review, meta-analysis and intervention content-analysis	Qualitative; Participative research process workshops	Mixed-methods: Think-aloud groups; participative research process workshops and online user- testing	Quantitative Feasibility RCT
MRC guidelines / Intervention mapping	Phase 0 / Step 1-6	Phase I / Step 1-4	Phase I / Step 1-6	Phase I / Step 1-6

^a Web-based self-management website including communication components and integrated online diary

Chapter 2 Methodology

2.1 Chapter overview

This chapter outlines the theoretical and methodological approach undertaken and presented in this thesis. It is outlined in two sections. The first section presents an overview of the intervention development frameworks used to generate research questions and guide intervention development including the process of translating the empirical literature and end-user feedback in intervention website design. The second section illustrates the joint application of the Medical Research Council (MRC) and intervention mapping (IM) guidelines for intervention development. The aims and rationale for each study are presented along with a description of the methods used.

2.1.1 Intervention development frameworks

2.1.2 2008 MRC Guidelines

The development of the online *Feeling Better* intervention was informed by the new Medical Research Council (MRC) guidance for the development of complex interventions (Craig et al., 2008). Complex interventions are interventions that have several interacting components or dimensions of complexity e.g. multiple components, number of behaviours targeted, the nature of the intervention outcomes, the variability in the target population and the degree of difficulty experienced by the individuals performing those behaviours. The purpose of these guidelines is to help researchers choose and apply appropriate methods within the context of their research (Craig et al., 2008; 2013). The updated framework accepts that each phase of research may be approached iteratively and not necessarily in a linear or even cyclical sequence. It is more important that within each phase of the research process the appropriate stages of planning and development are followed. The revised framework places greater emphasis on research piloting and early consideration of implementation issues which might improve the intervention and pre-empt obstacles prior to a final evaluation (Craig et al., 2008). As others have observed, the quality

Methodology of reporting of complex interventions has improved since the 2008 publication, however, these guidelines continue to be criticized as useful but too general for their purpose (French et al., 2012; Craig et al., 2013; Morrison et al., 2014a; Mohler, Kopke, & Meyer, 2015) (see Figure.2.1).

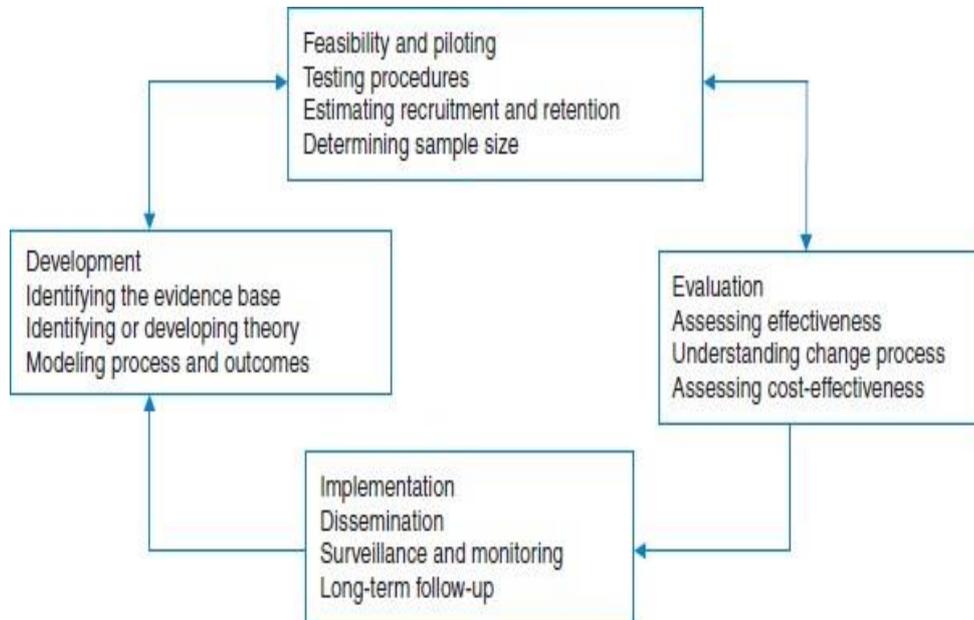


Figure 2.1 MRC framework for the evaluation of complex interventions (Craig et al., 2008)

The 2008 MRC guidelines lack detailed instruction on how to apply the various aspects of the framework (French et al., 2012). Examples of how the MRC guidelines may be implemented in practice all seem to share this limitation (Lakshman et al., 2014; Aventin, Lohan, O’Halloran & Henderson, 2015). This has inspired the development of multiple intervention development and implementation models which claim to build on the MRC guidelines (French et al., 2012).

2.1.3 Intervention Mapping

This study also employed the Intervention mapping (IM) protocol developed by Bartholomew, Parcel and Kok (1998). Intervention mapping is a detailed, stepped approach to theory driven, evidence-based intervention development. IM builds on the MRC guidelines by outlining

six steps to intervention development in which specific tasks must be completed to progress from problem identification to programme evaluation (Bartholomew, Parcel, & Kok, 1998) (see Figure 2.2). This approach complements the MRC framework by offering more detailed, incremental guidance on how each step in this process might be achieved (Table 2.1). The IM protocol has been used to develop interventions to promote healthy nutrition and physical activity in adolescents (Prins, van Empelen, Beenackers, Brug, & Oenema, 2010; Krølner et al., 2012); to support pain self-management for adults with chronic low back pain (Hurley et al., 2016); to inform an eHealth programme supporting adults returning to work post-surgery (Vonk Noordegraaf et al., 2012) and to successfully design disease prevention interventions (Garba & Gadanya, 2017). Published examples of IM vary in terms of the specificity with which they apply or report each step of the IM process. Therefore, the current application of the IM protocol emulates that of a similar intervention designed to promote pain management in adults with chronic low back pain (Hurley et al., 2016). The systematic approach employed by Hurley and colleagues is effective in communicating the many nuances of each step within the protocol and is adopted here.

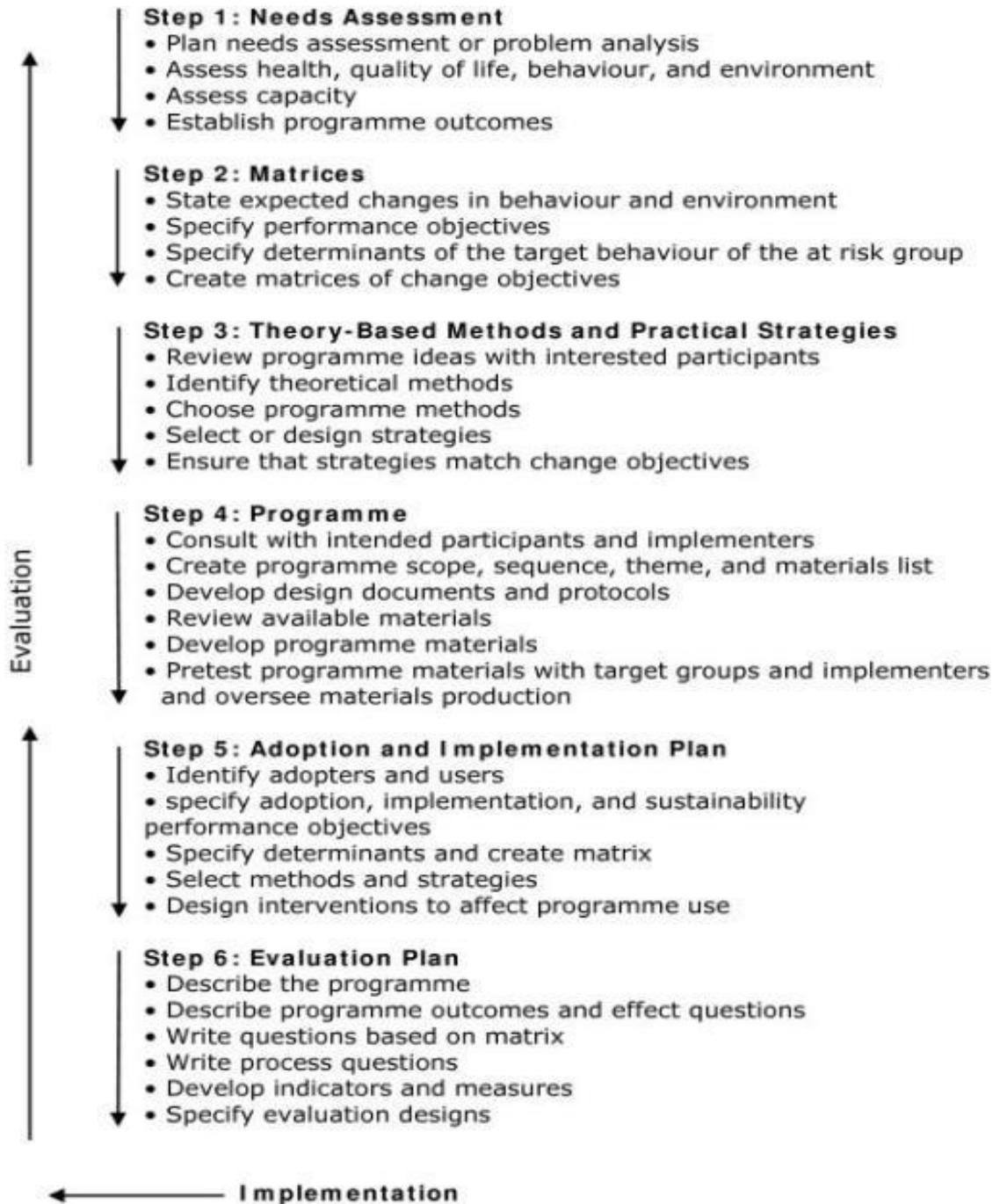


Figure 2.2 Intervention Mapping Protocol (Bartholomew et al., 1998)

Together these frameworks were used to provide a comprehensive account of intervention content, application of theory and hypothesized causal mechanisms (Table 2.1 *Studies undertaken mapped to the phases of the Medical Research Council / Intervention Mapping framework*). This process is outlined below:

Table 2.1

Studies undertaken mapped to the phases of the Medical Research Council / Intervention Mapping frameworks

Definition	Studies undertaken
1) Developing a complex intervention	
1.1 Identifying the evidence base	
Step 1. Needs assessment	(i) Scoping review of pain literature on the nature, impact and treatment of chronic pain and the experiences of pain self-management from the perspective of children, care-givers and health professionals to improve understanding of chronic pain self-management and the determinants of engagement in self-management practice. (ii) Systematically reviewed the empirical evidence for the effectiveness of ICT-based psychological interventions for paediatric pain management
1.2 Identifying/developing appropriate theory	
Step 1. Needs assessment	(i) Conducted qualitative study with child and parent stakeholders to validate evidence-based recommendations and refine intervention content. This involved participative research process workshops with children and parents. (ii) Optimise intervention design in line with end-user and expert feedback. (iii) Combined literature review, qualitative findings and expert feedback to select a theory, cognitive behavioural strategies, behaviour change techniques and modes of delivery.
Step 2 Matrices	
Step 3 Theory based methods and practical strategies	
1.3 Modelling process and outcomes	

Step 2 Matrices. Step 3 Theory based methods and practical strategies. Step 4 Programme

(i) Linked evidence and social cognitive theory based behavioural determinants to performance objectives (desired behaviour) and subsequently to behaviour change objectives (intervention targets), feasibility and clinical outcomes.

(ii) Selected evidence-based, validated questionnaires to assess change in social cognitive theory constructs and behaviour targeted by the intervention. (iii) Designed and developed programme and materials, pre-tested materials with end-users

2) Assessing feasibility and piloting methods

2.1 Testing procedures

Step 4 Programme

Step 5 Identify adoption and implementation plan

(i) Tested components for usability and acceptability and to inform final adaptation of the intervention,

(ii) Conducted 1 year, feasibility trial of the *Feeling Better* intervention (Jan 2016-Jan2017).

2.2 Estimating recruitment and retention

Step 4 Programme

Step 5 Identify adoption and implementation plan

Step 6 Evaluation plan

(i) Recruitment through the national media, social media, charities and parent-led support networks, (ii) Conduct feasibility trial (Jan 2016-Jan2017).

2.3 Determining sample size

Step 4 Programme

Feasibility trial was too small. Previous trials in this area were used to estimate sample size.

Step 5 Identify adoption and
implementation plan

Step 6 Evaluation plan

3) Evaluating a complex intervention

2.2 Research questions

2.2.1 Deductive approach – what do we already know?

One of the few, specific methodological recommendations made by the 2008 MRC framework is the conduct of a systematic review of the existing evidence base. The first step of the IM protocol expands this requirement to include a comprehensive needs assessment combining a review of the evidence base and appraisal of needs and resources for intervention development, adoption and implementation. Together these guidelines were used to generate the first research question: **Research question 1 (RQ1):** *What is the effectiveness of psychological therapies delivered using ICT in support of chronic pain self-management among pre-adolescent children with chronic pain and their care-givers?*

2.2.2 Inductive approach – what can we learn for ourselves?

The 2008 MRC guidelines next recommend the identification or development of a guiding theoretical framework which would allow greater understanding of the likely processes of behaviour change (Craig et al., 2008). This is in line with Step 2 of the IM protocol which requires careful planning of intervention objectives and desired outcomes in terms of the desired behaviour change. This process is conceptually applied using a matrix of performance and change objectives which functions as a map guiding the research team in the identification and application of an appropriate theoretical framework. Step 3 of the IM protocol is concerned with the practical application of theory and theoretical methods to better understand the determinants of behaviour change. Here the intervention matrix informs the selection of key therapeutic strategies, behaviour change techniques and intervention components which are consistent with the theoretical model and the match change objectives. These guidelines were used to generate the following research question: **Research question 2:** *What are the experiences, barriers and facilitators to paediatric chronic pain self-management from the perspective of the child with chronic pain and their parents or care-givers?*

2.2.3 Combined approach – do our findings build on what we know?

The MRC guidelines suggest a series of studies and designs might be necessary to appropriately model process and outcomes prior to full scale evaluation. IM Steps 4 to 6 expand on this with guided and incremental recommendations for iterative intervention development involving intervention adopters and users (Bartholomew et al., 1998). This is consistent with a person-centred approach to intervention development that is increasingly associated with greater intervention satisfaction, acceptability and adherence (Morrison et al., 2012; Yardley, Morrison, Bradbury, & Muller, 2015). These guidelines were used to generate a research question consistent with this approach: **Research question 3:** *How do younger children with chronic pain (5-12 years) and their care-givers perceive ICT-based therapy delivery and what are the design features and intervention components most likely to engage this population?*

2.2.4 Inductive - Is this worth pursuing?

The 2008 MRC guidelines recommend sufficient feasibility and piloting to anticipate and address issues such as poor recruitment, high attrition or smaller than expected effect sizes. In line with this, IM steps 4-6 require the development of an adoption and implementation plan. This is designed to avoid waste and duplication of research efforts or resources and encourage a complete understanding of the context in which the research is carried out such as the environment, social or cultural norms that might influence how the intervention is used. The final research question was generated to address this: **Research question 4:** *Is it feasible to develop and conduct a randomised controlled trial of an Internet-based version of the Feeling Better programme and how would such a programme be used by families coping with chronic pain?*

2.3 Application of guidelines

2.3.1 Step 1: Needs assessment

The first step in the development process was a literature search of Medline to understand the individual and environmental determinants of chronic pain and subsequent adjustment. The end-product was a scoping review of the empirical literature relating to psychological aspects of chronic pain in children and influencing factors that shape the behavioural and psychological response to pain. This summary of the literature formed the basis of Chapter 1. As a supplement to this literature search, child and parent needs, beliefs and treatment response were explored via three activities: (i) a systematic review of the empirical literature on the effectiveness of ICT-based psychological therapies for paediatric chronic pain, (ii) primary qualitative research exploring the lived experience of chronic pain self-management and theoretical analysis of this experience (iii) primary, mixed-methods research exploring the usability and acceptability of the prototype pain management programme. The cumulative aim of each study is to identify modifiable psychological factors and components that underpin effective pain management interventions and to inform the adaptation and development of an online version of the *Feeling Better* pain management intervention programme.

Systematic Review (Study One): This study was designed to address RQ1 by conducting a systematic review of the empirical literature on the effectiveness of information and communication technology as a method of psychological treatment delivery in support of paediatric chronic pain management (Chapter 3). This aimed to establish the effectiveness of psychological therapies delivered using ICT and to identify effective components and characteristics of such interventions (Study 1). A detailed account of this research is presented in Chapter 3.

Participative study of lived experience (Study Two): The purpose of this study was twofold. First to address RQ2 by conducting a primary, qualitative study of paediatric pain management from the perspective of pre-adolescent children and their care-givers. This was intended to supplement

the preceding review of the literature and gain insight into the lived experience of pain management from the perspective of a relatively neglected sub-group of the pain population. Second to ascertain clinical content and usability requirements of the prospective online FB intervention that might inform the development process. Findings from this study were retrospectively mapped to a theoretical framework (Social Cognitive Theory) (Bandura, 1996; 2005) and used to guide the development process.

Participant experiences, needs and preferences were examined in two ways: (i) by exploring participant perceptions of pain management in terms of what helps (facilitators), what do not help (barriers) and what we need to do better (improvement/solutions) and (ii) by brainstorming preferences for content and features that may be required of an intervention designed to promote pain self-management. Results pertaining to the former are presented in Chapter 3 whereas findings relating to intervention and website development are presented in the Chapter 4. This qualitative study (Study 2) involved two participative qualitative research groups of consenting children (n= 11) with various forms of juvenile idiopathic arthritis (JIA) and two participative groups of their care-givers (n= 21). Data analysis was both indicative and deductive. Inductive analyses were carried out by the consenting participants as part of the participative research process (PRP) approach. Deductive theoretical thematic analysis was carried out by the researchers (A.T. and SOH), using Social Cognitive Theory as a coding frame (Braun & Clarke, 2006). A detailed account of this research is presented in Chapter 4.

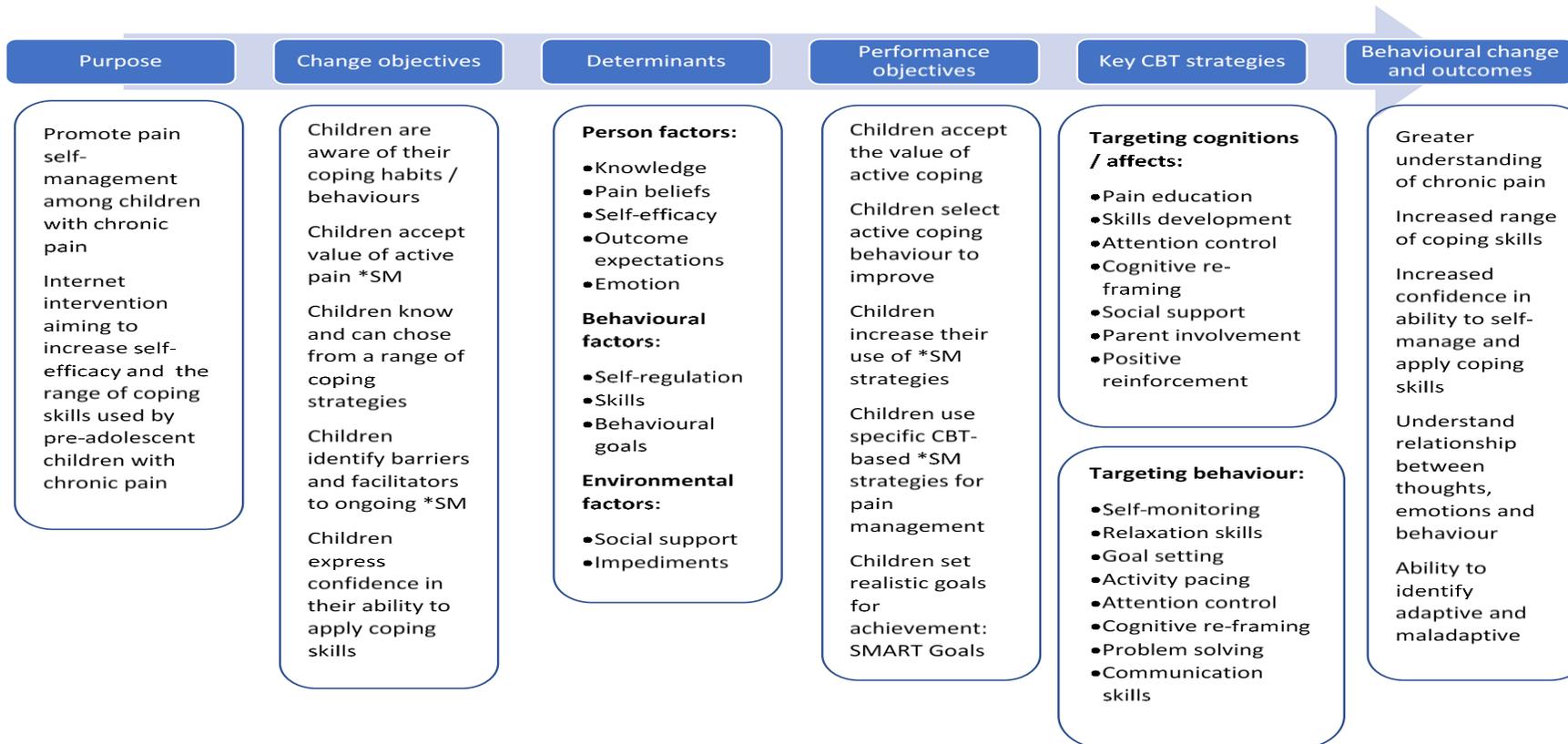
Mixed methods study of usability and acceptance (Study Three):

This study was designed to address RQ3 by exploring the usability and acceptability of each iteration of the online Feeling Better prototype and identify potential barriers and enablers of implementation and uptake by participants (Study 3). This involved an iterative and phased design and development process largely informed by end-user feedback in response to early and later drafts of the web-based prototype. The first phase of development was informed by think-aloud groups of consenting children (n=11) and their parents (n=21). The next iteration of the programme was

evaluated and refined in response to feedback from consenting children (n=5) with JIA and their parents (n=5) in a PRP group workshop. Final iterations of the programme were informed by consenting children (n=8) with juvenile idiopathic arthritis (JIA) or Ehlers-Danlos syndrome (EDS) and parents (n=8) in a staggered, pre-post, online user-testing of the web-based platform. Participant feedback was analysed according to the nine components outlined in Ritterband's Behaviour Change Model for Internet Interventions (Ritterband et al., 2009). This phase produced work-package (WP3), A detailed account of this research presented in Chapter 5.

2.3.2 Step 2: Matrices

Step 2 was concerned with the creation of a matrix which illustrates the causal relationship between the desired behavioural or environmental outcomes and the performance and change objectives required to achieve these outcomes (see Appendix 2). This is a record of how the desired outcomes (overall goals) of the *Feeling Better* intervention might link to the specific actions required to accomplish change in these outcomes (performance objectives/sub-behaviours) and to the change that needs to occur in the participant (change objectives) to accomplish these sub-behaviours. The desired change in the target population (i.e. children and parents) is specified in terms of the determinants of pain behaviour (e.g. elevated self-efficacy, lower pain catastrophising) that likely influence the performance of sub-behaviours (e.g. use of CBT coping strategies) (Bartholomew et al., 1998). Figure 2.3 represents a synthesis of the IM process in the context of chronic pain management. This is a condensed, visual summary of the *Feeling Better* matrix (Appendix 3).



*SM: self-management

Figure 2.3 Intervention Mapping: synthesis of intervention matrix

The evidence collected during the preceding needs assessment helped to identify the behavioural outcomes to be achieved and the selection of performance objectives (i.e. what the participant must learn, do or change to achieve that behavioural outcome) (Bartholomew et al., 1998). Performance objectives are very simply, specific actions required to accomplish change in the targeted behavioural and environmental outcomes. In this study, the intervention is designed to target modifiable, complex behaviour i.e. active coping behaviour. Pain self-management is an under-used coping approach in chronic pain populations. This is typically due to pain-related beliefs and the experience of impediments, personal or environmental. This may be addressed by breaking the complex behaviour down into smaller specific actions which may be shaped and/or measured. For example, this study defined the behaviour change as: (i) 'increased confidence in ability to self- manage and (ii) increased use of adaptive coping skills. This might be demonstrated through self and parent report of improvement in self-efficacy measured using the self-efficacy for functioning despite pain scale (Bursch, Tsao, Meldrum, & Zeltzer, 2006) and participant use of active coping strategies measured using the Pediatric Quality of Life – Pediatric Pain Coping Inventory (Varni et al., 1996) and objectively collected intervention usage data. Table 2.2 outlines the constructs and participant outcomes we planned to measure in the *Feeling Better* trial to better understand the hypothesised causal pathway (mediating mechanisms of behaviour change).

2.3.3 Step 3: Theory-based methods and practical strategies

The aim of Step 3 was to transform the change objectives to create and implement practical components within the intervention. This process was also informed by the preceding literature review and needs assessment which involved input from design team, potential end-users and experts in intervention development (Chapters 3, 4 and 5). The first task to be completed was the application of a theoretical framework, Social Cognitive Theory (Bandura, 2005) that might be combined with the determinants of adjustment to chronic pain identified in the initial literature review. SCT

identifies theoretical constructs that map to the proposed causal determinants of adaptive coping behaviour. These constructs may be targeted to using cognitive behavioural strategies and behaviour change techniques most likely to achieve the desired behaviour change. Behaviour change techniques (Michie et al., 2013) were implemented using various mode of delivery components (Webb et al., 2010) suited to the online environment in which the intervention is delivered and to the developmental level of the target population. The intervention prototype was independently coded for the presence or absence of the specified BCTs by two researchers (A.T. and E.M.).

2.3.4 Step 4: Programme plan

The aim of Step 4 was to create a programme plan and further refine the prototype of the intervention. Experts in intervention development and clinicians working with paediatric chronic pain patients offered informal assessments of the online prototype (n=4 intervention development experts and clinicians and n=2 clinical psychologists) and independently provided feedback on intervention content and usability. Adaptation of the *Feeling Better* CBT manual was conducted iteratively, in response to end-user and expert feedback. The existing *Feeling Better* manual is supported by published efficacy data using this CBT programme (McManus & McGuire, 2010; McManus et al., 2014).

The translation of clinical content in the *Feeling Better* manual to the proposed intervention website was guided by the checklist produced by the National Institutes of Health (NIH's) for the development of effective print materials for low-literacy audiences (NIH, 1998). The NIH Checklist outlines effective communication principles in terms of content/style, layout, visuals and readability. Literacy adaptation was carried out for all *Feeling Better* treatment modules and iteratively refined following each phase of user-testing. Content such as patient narratives, instructions on how to perform specific CBT strategies, client summary information sheets and educational materials relating to pain self-management were adapted to the

appropriate reading level and simplified where possible using visual illustrations. To reduce the level of intensity and adhere to an eight-week programme format, some of the CBT strategies in the *Feeling Better* manual were excluded from the final iteration of the online programme. The selection process was informed by SCT and the matrix of outcomes and objectives guiding this research. Selection was systematized using the MoSCoW Prioritisation method. The contents of the *Feeling Better* manual were categorised into groups of strategies and information labelled: Must have, Should have, Could have and Won't have time. Each page of the *Feeling Better* manual was labelled accordingly. Two authors (AT, BMG) reviewed the proposed intervention and decided on the core CBT components.

Table 2.2

Child and parent outcomes measured to assess the hypothesised mechanisms of behaviour change

SCT ^a domains of influence	Outcome	Description	Instrument
	Participant recruitment / attrition	Recruitment over time Reasons for non-participation Reasons for drop out	System usage data and tailored data collection forms
	Integrity of study protocol	Technical functioning Proportion of completed measures Child and parent adherence	
	Acceptability	Ease of use, relevance, ease of understanding, satisfaction	Internet Evaluation and Utility Questionnaire (Thorndike et al., 2008)
		Adaptation	6 purposely constructed open-ended questions

Person factors (beliefs, appraisals biologic events)	Estimates of treatment effect	Functioning: physical, social, emotional and role	Pediatric Quality of Life Inventory -(PedsQL™ 4.0; Varni, 1998)*.
		Pain intensity	Wong-Baker FACES Pain Rating Scale*
Self-efficacy		Self-efficacy	Self-efficacy for functioning despite chronic pain (Bursch et. al., 2006).
		Pain Catastrophising	Pain Catastrophising Scale-child and parent versions (PCS- C&P; Crombez et al., 2003).
Outcome expectations/ barriers			
Behaviour factors (skills, habits)	Use of coping strategies	Coping skills	Pediatric Quality of Life Inventory – Coping Skills Inventory (PedQL-CSI; Varni, 1998).
Environmental factors (social support, facilitation, barriers)	Parental protectiveness	Protective behaviour	Adult Response to Children’s Symptoms–Protect Subscale- child and parent versions (Walker, Ley & Whitehead, 2006).

^a Social Cognitive Theory, *based on McGrath et al., (2008) PedIMMPACT Recommendations

2.3.5 Step five: Adoption and implementation plan

As per the 2008 MRC guidance, implementation issues were considered early in the intervention development process. IM requires that programme-use outcomes are identified that might demonstrate successful adoption and implementation of the programme by participants. This selection was informed by the preceding needs assessment and review of the literature. The overall goal of this intervention was to promote increased active coping behaviour among pre-adolescent children with chronic pain. Thus, it was decided adoption would be best demonstrated through regular use of the intervention defined as 10-30 minutes of active coping behaviour on a weekly basis. This outcome was practically applied by incorporating it in the intervention's weekly prescriptions for pain self-management via weekly check-in and check-out, homework compliance, streaming of instructional video or audio files and completion of interactive exercises (CBT strategies) and self-monitoring activities. This process was facilitated using selected evidence-based BCTs and modes of delivery (i.e. design features).

2.3.6 Step six: Evaluation Plan

As might be expected, the evaluation plan for the online Feeling Better intervention is focused on feasibility and piloting that we might draw conclusions on the feasibility and potential effectiveness of the proposed intervention. The primary outcomes of interest are feasibility outcomes relating to recruitment, adherence, retention and treatment satisfaction which might inform a definitive trial. Clinical outcomes of interest are those recommended by the PedIMMPACT consensus statement including physical, social and emotional functioning and pain intensity (McGrath et al., 2008). The pilot trial (Jan 2016–Jan 2017) and all associated recruitment, conduct and assessment procedures was carried out online. A total of 67 child-parent dyads were randomised to one of two arms (Internet intervention or Waitlist control). Children and parents were assessed at three

points and completed treatment satisfaction questionnaires. It is expected these findings will inform the design and delivery of a definitive, larger trial.

2.3.7 Conclusion

This chapter outlined the planned development of a complex intervention using an intervention mapping protocol. The results of this process are described in each of the succeeding chapters. Chapter 3, 4 and 5 adhere to Steps 1-4 of the IM protocol. Chapter 5 and 6 adhere to Steps 4-6 of the IM protocol. The utility of the IM protocol is discussed in Chapter 7.

Chapter 3 The effectiveness of information and communication technology-based interventions for paediatric chronic pain: A systematic review, meta-analysis and intervention content analysis (Study 1)

3.1.1 Publication:

Traynor, Morrissey, Egan, & McGuire, (2016). The effectiveness of information and communication technology-based psychological interventions for paediatric chronic pain: Protocol for a systematic review, meta-analysis and intervention content analysis. *BMC Systematic Reviews*

3.1.2 Highlights

- A systematic review of 10 clinical trials investigating the effectiveness of technology-based psychological interventions
- Psychological therapy delivered using information and communication technology may be a promising and potentially effective treatment for children with chronic headache.
- We provide recommendations for future research

3.2 Chapter overview

This chapter presents the systematic review of existing evidence conducted as part of the need assessment outlined in Chapter II. The primary aim was to systematically review existing evidence for the use of psychological therapies delivered using information and communication technology to support chronic pain management among children and adolescents. A secondary aim of this research was to explore the content of existing treatment interventions that may inform the proposed intervention development. As previously reported, a three-fold approach was undertaken to achieve these aims. The first, a systematic review of the empirical literature, the second a meta-analysis of intervention components and the third, a retrospective content analysis using established intervention development frameworks. The findings from this study informed the next step in the research process and identified gaps in the pain literature that might be addressed by the present research.

3.3 Introduction

Following our initial review of the paediatric pain literature (Chapter 1) we decided to focus one potential solution to access and resource issues which prevent the uptake of psychological therapies in support of paediatric pain management. Systematic evaluations of psychological therapies are increasingly including information and communication technology (ICT)-based trials. Research in this field is expanding quickly to address barriers to accessing psychological therapy. The evidence base supporting the use ICT-based therapies as a source of support for paediatric pain management is inconclusive.

Previous systematic reviews and meta-analyses of technology-based psychological interventions for paediatric chronic pain have been performed (Stinson et al., 2009; Macea et al., 2010; Fisher et al., 2015). Stinson et al (2009) undertook a review of Internet delivered interventions for paediatric chronic health conditions. The review included nine randomised controlled trials of which only one focused on chronic pain patients (Hicks et al., 2006;

also included in this review). The authors were unable to conduct meta-analyses due to the heterogeneity of included conditions and measures. Macea et al., (2010) investigated Internet based cognitive behavioural therapy (CBT) interventions for adults and children with chronic pain. Eleven randomised controlled trials (N = 2953) were identified, only two of which focused on children and are included in the present review (Connelly, Rapoff, Thompson, & Connelly, 2006; Hicks et al., 2006). Small reductions in pain were observed in response to the web-based treatment interventions. Other core outcomes such as disability or emotional distress were not investigated. Fisher et al., (2015) was the first and only review to date which focuses solely on the efficacy of remotely delivered interventions for chronic pain in children and young people. The authors included eight randomised controlled trials (N=371 participants) and like the current review analysed headache and mixed pain conditions separately. Significant beneficial effects were observed for headache severity and pain intensity among children with mixed pain conditions at post-treatment but not at follow-up. No other beneficial effects were observed, and the authors were prevented from conducting follow-up analyses of disability and depression across pain conditions due to insufficient data.

These previous systematic reviews and meta-analyses were limited in several ways. The search strategies employed were not designed to search for unpublished controlled trials. Some analyses were prevented due to insufficient data. The effectiveness of the included trials varied considerably, and intervention content was not considered in terms of the components which may be contributing to estimates of effect. It was therefore difficult to discern the active ingredients in the interventions or their mechanisms of action. Contributing to current understanding in this area would be beneficial for intervention development teams working on self-management programmes for young people with chronic conditions.

Existing systematic reviews and meta-analyses of technology-based psychological interventions for paediatric chronic pain have provided preliminary support for the use of remotely delivered psychological therapy for paediatric pain management (Fisher, Law, Palermo, & Eccleston, 2015;

Macea, Gajos, Daglia Calil, & Fregni, 2010; Stinson et al., 2009). However, in the years since their publication, additional treatment evaluation trials have emerged that may have important implications for this field.

Chronic pain is a complex problem which is typically addressed using complex, multi-factorial interventions. It is a commonly cited limitation that such interventions do not report the explicit use of theory (Michie & Johnson, 2012). Furthermore, even when interventions are described as theory-based they may fail to report how or why theory was used in intervention development, content or recruitment (Little, Preece, & Eccles, 2015). According to the Medical Research Council guidelines for the development of complex interventions, theory-informed interventions are associated with greater efficacy than non-theory-based studies (Craig et al., 2013). Since the publication of the Medical Research Council guidelines for the development of complex health interventions, there has been a greater focus on identifying the circumstances in which a given intervention is most likely to be effective.

Recent studies have successfully attempted to synthesise existing evidence retrospectively (Little et al., 2015). A review by Webb et al., (2010) considered the relationship between intervention content (theoretical basis, behaviour change techniques (BCTs) and modes of delivery) and the effectiveness of Internet-based interventions for health behaviour change. The review, containing 85 controlled trials (N = 43,236) observed that extensive use of theory was associated with increases in effect size. Interventions using a greater number of BCTs tended to have larger effects and effectiveness was enhanced in studies which used additional methods of communicating with participants. However, this review only included adult-focused interventions and did not evaluate interventions for chronic pain management (Webb et al., 2010). The authors concluded that future research should compare intervention components including the effects of behaviour change techniques and intervention usability as potential sources of variability in estimates of beneficial effect. This is particularly important given the significant time and resources typically allocated to ICT-based intervention development and content adaptation. The use of established

theoretical and behavioural frameworks such as the Theoretical Domains Framework (Cane, O'Connor, & Michie, 2012), the behaviour change taxonomy (Michie et al., 2013) and novel functionality coding schemes (Webb et al., 2010) facilitate accurate description of behavioural interventions and a shared language for intervention development. To the best of our knowledge, content analyses of ICT-based psychological interventions for paediatric pain have not been performed, nor have behaviour change frameworks been used to retrospectively evaluate randomised controlled trials for paediatric pain conditions.

3.3.1 Focus of the Systematic Review and Meta-Analysis

The aims of the current systematic review and meta-analysis were as follows:

- To determine the overall effect of psychological therapies delivered using information and communication technology on pain intensity, pain-related disability, psychological distress (anxiety and depression), quality of life and treatment satisfaction at post-treatment and follow-up among children and adolescents with chronic pain
- To identify the theoretical basis, behaviour change techniques, mode of delivery, intensity and usability factors that may be associated with estimates of effect in ICT-based interventions.

We attempted to make several methodological advances in this review, as follows:

We have included randomised controlled trials (RCTs) that have been complete since the publication of previous reviews.

We have expanded outcome assessment to include quantitative assessment of quality of life and treatment satisfaction. Previous reviews have included quality of life scores in analyses of the effects of therapy on disability, such that quality of life was synonymous with disability. We have

chosen to look at disability and quality of life as distinct concepts that may produce distinct effects in response to psychological treatments for chronic pain. The addition of treatment satisfaction in meta- analyses may provide important information about the acceptability of technology-based therapies.

We separately examined study characteristics that may influence outcomes such as (i) the nature of the control condition (e.g. waitlist, minimal attention control and active control conditions), (ii) the point at which participants are lost to follow-up (pre-enrolment, post-treatment and follow-up assessment), and (iii) the analytic approach used (e.g. ‘as- treated’ or ‘intent to treat’ analyses).

We used established coding frameworks to (i) systematically categorise intervention content in terms of its theoretical basis (using the Theoretical Domains Framework; Cane et al., 2012); (ii) describe the extent to which there were identifiable behaviour change techniques with the treatments (using the behaviour change taxonomy version 1; BCTTv1; Michie et al., 2013) and (iii) to identify the modes of delivery used for the therapies (using a novel mode of delivery coding scheme; Webb et al., 2010).

3.4 Methods

3.4.1 Methodological framework

The Preferred Reporting Items for Systematic Review and Meta-Analysis (PRISMA; Moher, et al., 2009) statement was used to develop the current methodological framework and associated protocol. This review is registered with the International Prospective Register of Systematic Reviews (PROSPERO) database (registration number: [CRD42016017657](#)). The associated protocol has been published and may be viewed in Appendix 4 (Traynor, Morrissey, Egan, & McGuire, 2016).

3.4.2 Eligibility criteria:

3.4.3 Types of participants

Studies were included if they focused on children and adolescents under 18 years of age, diagnosed with non-malignant, chronic or persistent pain or who have experienced chronic pain for a period of three months or more.

3.4.4 Types of intervention

Studies were included if one arm of each trial was of a psychological therapy delivered using information and communication technology as the dominant mode of delivery. Studies were included if they evaluated self-led information and communication technology-based psychosocial therapeutic interventions. Studies must have used information and communication technology (ICT) as the primary source of intervention delivery. Studies had to include an arm of a behavioural and/or psychological intervention with definable psychotherapeutic content targeting chronic pain management. Interventions that provided information/education only, without a therapeutic component or social support were excluded.

3.4.5 Types of comparisons

Studies had to include a control condition including usual care, waiting list control or an active control condition.

3.4.6 Types of outcomes

Studies were included in the meta-analysis if they reported statistical data allowing the calculation of effect sizes on at least one pain-related outcome obtained from the article or provided by the author(s) upon request. Outcomes of interest included pain intensity, pain interference, emotional distress (depression and/or anxiety) and quality of life measured at baseline (pre-treatment), at post-treatment and/or at follow-up.

3.4.7 Types of studies

Studies were included if they were randomised or cluster-randomised controlled trials, published or unpublished and reported in English.

3.4.8 Identification of studies

We searched the following electronic bibliographic databases from their inception through to May 2015: OVID: Embase (1980 (database inception) to May 2016), MEDLINE (1980-May 2016), PsycINFO (1980 to May 2016), and CENTAL (1999 to May 2016). This search strategy included a search for unpublished randomised controlled trials across grey literature sources such as Scopus, Ethos, Trip, and across clinical trial registries including ClinicalTrials.gov (clinicaltrials.gov), the metaRegister of controlled trials (mRCT), (controlled-trials.com) and the World Health Organisation (WHO) International Clinical Trials Registry Platform (ICTRP) (www.who.int/ictip/en/) for trials. We concluded our search by scanning the reference lists of included studies and relevant systematic reviews. In addition, leading authors in the field of paediatric chronic pain management were contacted to identify relevant studies. The full search

criteria and keywords are available in Appendix 5 and in the published protocol (Traynor et al., 2016).

3.4.9 Data extraction and coding

Two reviewers (AT, BMG) independently screened titles, abstracts and full reports of studies which initially appeared to meet inclusion criteria. All screening decisions were cross verified, and full-text articles retrieved for further assessment of eligibility. Data extraction was carried out by the first author and checked by the last author. A data extraction form was developed and piloted on one included study (see Appendix 6). The following information was extracted from each of the included studies: bibliographic information (authors, year of publication, country, reference), condition-specific information (diagnosis, duration of pain), sample characteristics (sample size, gender, age), mode of delivery (computer, smartphone, telephone), mode of delivery characteristics (automated, communicative, supplemental, enriched, tailored), type of treatment (cognitive behavioural therapy, other behavioural and psychological approaches), type of care provided to the control group (passive control - waiting list control, active control - relaxation), outcomes assessed (pain intensity, pain interference, anxiety, depression, quality of life), outcome measurement tools used (type and name of measure), assessment points and time at which drop-out occurred (baseline, post-treatment, follow-up) and the analytic approach used (as treated or intention to treat). Inter-rater reliability was calculated prior to resolving discrepancies (Cohen's kappa: $\kappa = 0.872$, substantial agreement (Landis & Koch, 1977)). Discrepancies were discussed until 100% agreement was achieved.

Content analysis was performed using evidence from the text which included the author's own description of the intervention and the literature used to justify intervention content (e.g. table of contents). The theoretical basis of the included intervention and control conditions was assessed using the Theoretical Domains Framework (TDF) (Cane et al., 2012). The TDF domains that appeared to be targeted in the included intervention and

control conditions were identified and coded independently by two authors (AT and EM). Coding decisions were informed by the constructs and determinants which could be inferred from the text and by reference to the construct definitions set out by Cane et al., (2012). The coding of the theoretical domains targeted in the intervention and control groups for each of the studies is shown in detail in Appendix 7. In line with the analytic approach taken by Little et al., (2015) the relationship between the total number and frequency of different constructs and estimates of effect were explored separately in outcome analyses.

To identify the “active ingredients” or behaviour change techniques potentially contributing to intervention effectiveness, we assessed the strategies used to promote adaptive coping with pain and/or improvement in pain related knowledge and beliefs. The content of the included intervention and control conditions were coded independently by two authors (AT and EM), using the Behaviour Change Taxonomy v1 (Michie et al., 2013) and a data extraction form designed for the purpose (see Appendix 8). The data extraction form was tested on three randomly selected included studies. Two authors, (AT, EM) independently coded the following characteristics: i) the presence vs absence of behaviour change techniques and ii) the total number BCT's identified in the selected experimental and control conditions and iii) the frequency of different of different BCT's identified in the selected experimental and control conditions. Disagreements in coding were resolved by discussion. The influence of BCTs on estimates of effect was explored separately in subgroup analyses. Inter-rater reliability was calculated prior to resolving discrepancies (Cohen's kappa: $\kappa = 0.758$, indicating substantial agreement (Landis & Koch, 1977)). Discrepancies were discussed until 100% agreement was achieved.

To identify the mode of delivery that potentially contributed to intervention effectiveness, we evaluated mode of delivery in terms of usability. Using a modified version of the coding scheme developed by Webb et al., (2010), mode of delivery was assessed according to the following categories (i) automated functions, (ii) communicative functions,

and (iii) use of supplementary, (iv) navigational format (v) entertainment (iv) credibility modes (i.e. confidence in the accuracy of the information provided). A complete list of delivery modes can be seen in Table 3.1.

Table 3.1

Modified mode of delivery coding scheme (Webb et al., 2010)

Mode of delivery: Automated functions
(a) Enriched information environment (e.g. supplementary content and links, testimonials, videos, tapes, games etc.)
(b) Automated tailored feedback based on individual progress monitoring (e.g. comparison to goals, reinforcing messages, coping messages etc.)
(c) Generic follow-up messages (e.g. reminders, tips, newsletters, encouragement)
(d) Tailored initial advice (on basis of answers to questions about beliefs, problems, circumstances).
(e) Supported progress monitoring (e.g. electronic diary/chart).
Mode of delivery: Communicative functions (tick all that apply)
(f) Access to advisor to request advice (e.g. 'ask the expert' facility, expert-led discussion board or chat sessions)
(g) Scheduled contact with advisor (e.g. emails)
(h) Peer-to-peer access (e.g. buddy systems, peer-to-peer discussions boards/forums, live chat)
Supplementary modes of delivery:
(i) Email
(j) Telephone
(k) Text message (SMS)
(l) CD ROM
(m) video conferencing
(n) Web-based
(o) Other (e.g. instant-messaging)
Navigational format:
(p) tunneled: progress dependent upon completion of preceding module; free choice
(q) free choice (specify)
Entertainment value (enticing features):
(r) quizzes, stories, graphics (specify)
(s) appearance (e.g. colour, layout etc.) (specify)
Credibility (confidence in source):
(t) sources, credentials cited (specify)
(u) other specify

3.4.10 Quality evaluation

3.4.11 Risk of bias

Two reviewers (AT, BMG) independently assessed the risk of bias in the included studies using the Cochrane Collaboration tool for assessing risk of bias (Higgins et al., 2011). The following domains were rated: random sequence generation, allocation concealment, blinding of outcome assessment, incomplete outcome data, and selective reporting. The first and last authors rated all studies and cross-verified. There was perfect agreement between the two authors.

3.4.12 Methodological quality of results

The "Grades of Recommendation, Assessment, Development, and Evaluation" (GRADE) approach (Balshem, Helfand, Schunemann, Oxman, Kunz, Brozek, ... Guyatt, 2011; Guyatt, 2011; Guyatt et al., 2013) was used to evaluate the quality of results. Rather than assess individual studies, pooled data for each outcome is assessed and graded. Quality assessment is based on the following factors: ii) indirectness of evidence iii) inconsistency of results iv) imprecision of effect sizes and v) potential publication bias. Each outcome is graded based on confidence in the estimate of effect and ranked according to the following quality categories:

- High quality: further research is very unlikely to change our confidence in the estimate of effect.
- Moderate quality: further research is likely to have an important impact on our confidence in the estimate of effect and may change the estimate.
- Low quality: further research is very likely to have an important impact on our confidence in the estimate of effect and is likely to change the estimate.
- Very low quality: we are very uncertain about the estimate.

3.4.13 Data Analytic Strategy

Meta-analyses were conducted separately for each of the four dependent variables. We followed Eccleston et al (2014b) and Fisher et al (2015) by dividing and analysing included trial separately according to whether the experimental intervention was focused on headache or mixed pain conditions. Continuous outcome measures were assessed using standardised mean differences (SMDs) instead of mean scores to compensate for heterogeneity of extracted outcomes in most trials. For categorical data we used Mantel–Haenszel odds ratios (ORs) to assess the main categorical outcome measures. Effect sizes were calculated from intention-to-treat (ITT) data, or from complete cases data, if appropriate ITT data were not available. Planned sensitivity and subgroup analyses were used to explore the robustness of the results. To confirm the validity of the results obtained, primary analyses were repeated excluding studies presenting a high or uncertain risk of bias across categories. To identify the "active ingredients" contributing to effective interventions we also conducted subgroup analyses of intervention characteristics and content condition used (e.g. psychological intervention versus a passive or active control condition), the mode of delivery (e.g. automated, communicative, supplemental, navigational format, entertainment value and credibility) and the inclusion or exclusion of certain BCTs and TDF domains. Sensitivity and subgroup analyses were only performed if ≥ 2 studies were able to be pooled. A post-hoc subgroup analysis was conducted to evaluate the acceptability of ICT-based trials based on loss-to-follow-up. This was determined through comparing endpoint numbers of drop-outs from the intervention and control groups.

Data were pooled in random-effects meta-analysis in Review Manager 5.3. Due to the expected heterogeneity in intervention outcome measures a random effects model was used in all analyses (Higgins, 2011). Heterogeneity between studies was assessed using the Q statistic ($P < 0.10$) and I^2 index. Values of 0–40% were deemed to represent unimportant heterogeneity, 30%–60% represent moderate heterogeneity, 50%–90% represent substantial heterogeneity, and 75%–100% represent considerable heterogeneity (Higgins, 2011). Tests for funnel plot asymmetry were not

performed because fewer than ten trials were included in meta-analyses meaning the power of the test would be too low to distinguish chance of true asymmetry. Moderator analyses of effects could not be explored due to the small number of studies (<10 trials, <3 trials in most comparisons).

3.5 Results

3.5.1 Search results

The study selection process and reasons for exclusion are illustrated in Figure 3.1 using the PRISMA flow diagram (Moher, Liberati, Tetzlaff & Altman, 2009). The search produced 646 unique records. We excluded 626 records. The main reasons for exclusion were the intervention not being (i) a randomised controlled trial, (ii) psychological in content, (iii) not enough participants in each arm of the trial at post-treatment and (iv) ICT was not the primary mode of delivery. Remaining abstracts were assessed and a total of 20 papers were subject to full-report screening. A total of 10 papers fulfilled the inclusion criteria (see Tables 3.2 & 3.3). Of those, only 9 papers provided the necessary data for effect size calculation allowing inclusion in the meta-analysis (Table 3.8).

3.5.2 Study Characteristics

The characteristics of the 10 included studies are summarized in Table 3.2 and 3.3. The trials were published between 1992 and 2015. All but one trial (Trautmann & Kröner-Herwig, 2010) was conducted in either the United States or Canada. Sample sizes varied from 35 to 269 participants. Six trials investigated the effects of ICT-delivered psychological therapy for children with chronic headache (McGrath et al., 1992; Connelly et al., 2006; Cottrell, Drew, Gibson, Holroyd, & O'Donnell, 2007; Trautmann & Kröner-Herwig, 2010; Rapoff et al., 2014; Law, Beals-Erickson, Noel, Claar, & Palermo, 2015). One trial evaluated the effects of ICT-delivered psychological therapy for children with juvenile idiopathic arthritis (Stinson, 2010a). Three trials investigated the effects of ICT-delivered psychological therapy for children with headache and mixed pain conditions including recurrent abdominal pain, musculoskeletal pain and headache (Hicks et al., 2006; Palermo et al., 2009, Palermo et al., 2016). Data from these trials were entered in both headache and mixed pain analyses where appropriate. Eight trials relied on clinician referral and/or recruitment from participating hospitals and clinics (McGrath 1992;

Connelly et al., 2006; Cottrell et al., 2007; Palermo et al., 2009; Rapoff et al., 2014; Stinson, 2010a, Law et al., 2015, Palermo et al., 2016). One trial recruited from the community and through advertising in clinics (Hicks et al., 2006) and one trial only recruited from the community using various forms of media advertising (Trautmann & Kröner-Herwig, 2010).



PRISMA 2009 Flow Diagram

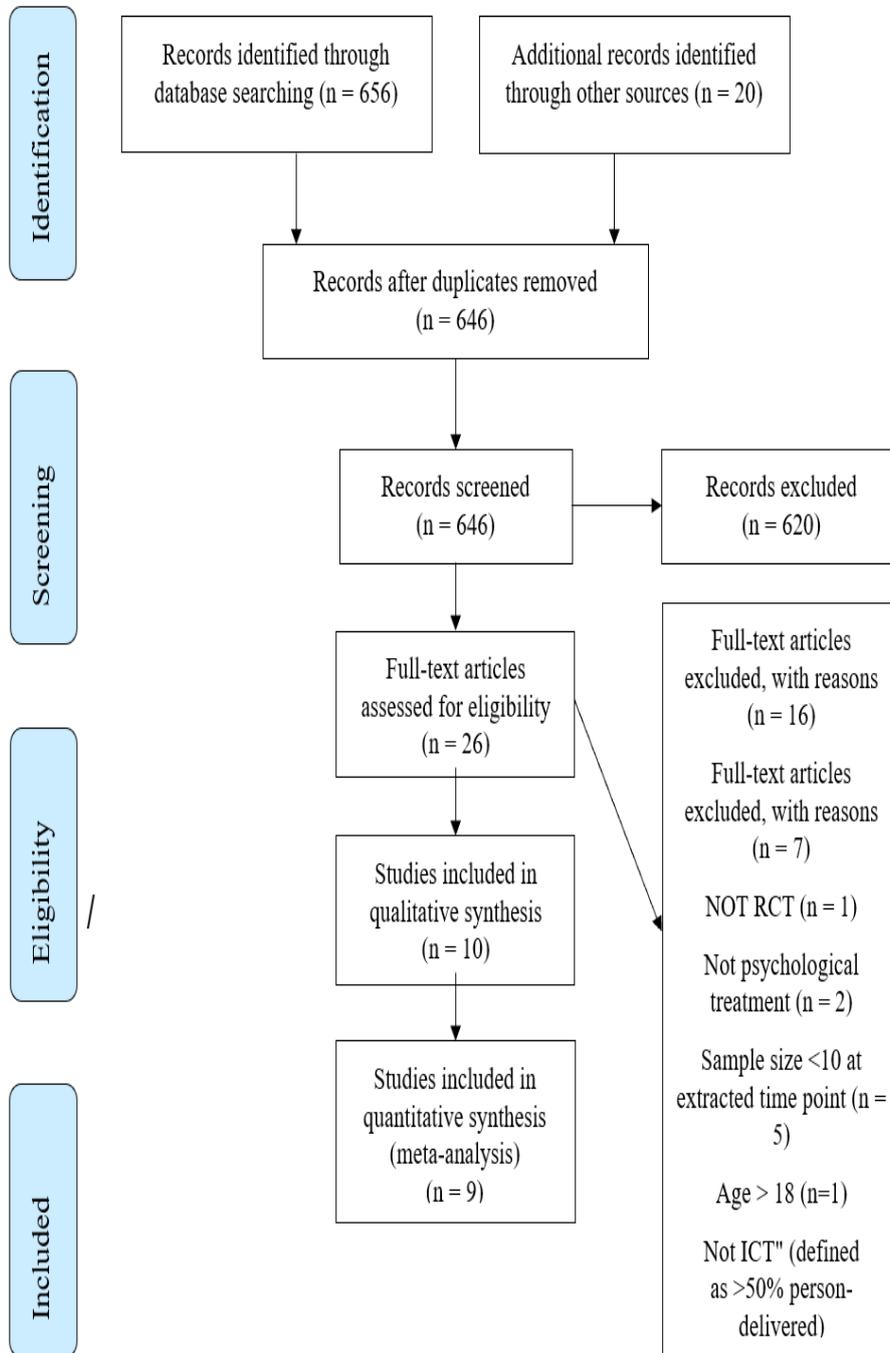


Figure 3.1 PRISMA (2009) flow diagram (Moher, Liberati, Tetzlaff & Altman, 2009).

3.6.1 Characteristics of participants

A total of 747 participants entered the treatment interventions and 623 completed giving a retention rate of 83.4%. Participant age ranged from 7-18 years. Participants included a total of 469 girls and 278 boys therefore, girls outnumbered boys by 60%. All included trials reported children had received a diagnosis of their pain condition by a medical professional.

3.6.2 Characteristics of the ICT-based Interventions

All the included experimental interventions either explicitly reported the use of a cognitive behavioural framework or described therapeutic content as consistent with cognitive behavioural therapy principles for pain management. Eight of the included trials compared two arms and two trials compared three arms (McGrath et al., 1992; Trautmann & Kröner-Herwig, 2010). As per the protocol, we compared the ICT-based intervention with the active control condition. For McGrath et al (1992) we compared the ICT-based intervention to the clinic-based control condition. For Trautmann & Kröner-Herwig (2010) we combined the active treatment conditions and compared this to the education control condition.

3.6.3 Characteristics of comparison groups

Two of the trials utilised waiting-list control groups for comparative data (Hicks et al., 2006; Palermo et al., 2009), two trials utilised attention control conditions consisting of weekly phone calls (Connelly et al., 2006; Stinson et al., 2010a). The remaining six trials report using an active education-based control condition, however only three of these trials used an active control condition which attempted to equalise treatment time, attention and mode of delivery (Trautmann & Kröner-Herwig, 2010; Rapoff et al., 2014 and Palermo et al., 2016).

Table 3.2

Characteristics of included studies

Study	Pain condition	Participants N (% female), age range (mean / SD)	Design and analysis	Recruitment (period)	Intervention (N)	Control (N)	Duration of treatment (mins)	Timing of outcome assessment	Withdrawal (intervention, control)
Connelly et al 2006, USA	Headache	N = 37 18F (49%), 7-12 yrs., (M = 9.9, SD = 1.66)	Pilot RCT, ITT	Clinic	CBT CD-ROM (n=17), 4 sessions / 4 weeks (60 mins),	Attention control (n = 20), weekly phone call	4 weeks (60 mins)	Baseline, post-treatment: 4 weeks, follow-up: 3 mos.	n=6/37, (CBT= 3/17, WLC= 3/20)
Cottrell et al., 2007, USA	Headache	N=30, 15F (50%), 12-17 yrs., (M = 14.1, SD = 1.91)	RCT, ITT	Clinic and community	Telephone administered treatment (n =18), 8 sessions / 8 weeks (240 mins)	Triptan treatment (n = 16),	8 weeks (240 mins)	Baseline, 3mos, follow-up: 8 mos.	n=6/34, (CBT= 4/18, WLC= 2/16)

Hicks et al., 2006, Canada	Recurrent paediatric headache, abdominal pain	N= 47, 30F (64%), 9-16 yrs.; (M = 11.7, SD = 2.1	RCT, ITT	Community, (1 yr.)	Internet based CBT (n = 25) 7 sessions /r 7 weeks, + email contact (NR mins)	Usual care WLC (n=22)	7 weeks (NR)	Baseline, post-treatment: 1 mo. Follow-up: 3 mos.	n=15/47, CBT= 7/25, WLC= 8/22)
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CBT: cognitive behaviour therapy; WLC: waitlist control; ITT: intent to treat; NR: not reported; N: total sample size; n: group sample size

Table 3.2
Characteristics of included studies continued

Study	Pain condition	Participants N (% female), age range (mean / SD)	Design and analysis	Recruitment (period)	Intervention (N)	Control (N)	Duration of treatment (mins)	Timing of outcome assessment	Withdrawal (intervention, control)
Law 2015, USA	Headache	N=83, 68F (81.9%), 11-17 yrs., (M = 14.5, SD = 1.7)	RCT, ITT	Headache clinic (2 yrs.)	Internet based CBT (n = 44), + email contact, 8 sessions / 8-10 weeks (240 mins)	Headache treatment clinic: medication management, psychological therapy (CBT/biofeedback/physical therapy) (n=39)	8-10 weeks (240 mins)	Baseline, 8-10 weeks, follow-up: 3 mos.	n=34/83, (CBT= 17/44, WLC= 17/39)
McGrath 1992, Canada	Headache	N=87, 63F (72%), 11-18 yrs. (NR)	RCT	Clinic	CBT Tapes, telephone calls and manual (n=21), 8 sessions / 8 weeks (240 mins)	CBT face to face (n= 20)	8 weeks (240 mins)	Baseline, post-treatment: 1 mo. 3 mos., 1 year	n=12/59, (CBT= 6/30, WLC= 6/29)

CBT: cognitive behaviour therapy; WLC: waitlist control; ITT: intent to treat; NR: not reported; N: total sample size; n: group sample size

Table 3.2

Characteristics of included studies continued

Study	Pain condition	Participants N (% female), age range (mean / SD)	Design and analysis	Recruitment (period)	Intervention (N)	Control (N)	Duration of treatment (mins)	Timing of outcome assessment	Withdrawal (intervention, control)
Palermo et al 2009, USA	Chronic idiopathic pain (abdominal, headache, musculoskeletal pain)	N = 48, 35F (72.9%),	RCT, ITT	Clinic (2 yrs.)	Internet based CBT group (n= 24), + email contact, 8 sessions / 8- 10 weeks (240)	WLC (n = 22),	8-10 weeks (240 mins)	Baseline, post- treatment: 8- 10 weeks, follow-up: 6 mos., (12 mos. ongoing)	n=2/48, (CBT=1/24, WLC=1/22)
Palermo et al 2016, USA	Chronic idiopathic pain	N=269, 205F (75.1%), 11-17 yrs., (M = 14.7, SD = 1.6)	RCT, ITT	Clinic (3.5 yrs.)	Internet based CBT group (n= 134), + email contact, 8 sessions / 8- 10 weeks (240 mins)	Internet education group, (n=135)	8-10 weeks (240 mins)	Baseline, post- treatment: 8- 10 weeks, follow-up: 6 mos.	n=9/269, (CBT=8/133, WLC=1/133)

CBT: cognitive behaviour therapy; WLC: waitlist control; ITT: intent to treat; NR: not reported; N: total sample size; n: group sample size

Table 3.2

Characteristics of included studies continued

Study	Pain condition	Participants N (% female), age range (mean / SD)	Design and analysis	Recruitment (period)	Intervention (N)	Control (N)	Duration of treatment (mins)	Timing of outcome assessment	Withdrawal (intervention, control)
Rapoff 2014, USA	Headache	N=35, 25F (71%), 7-12 yrs., (M = 10.2, SD = 1.7)	RCT	Clinic (5.3 yrs.)	CBT CD-ROM (n=18), 4 sessions /4 weeks (240 mins)	CD-ROM Educational module (n = 17),	4 weeks (60 mins)	Baseline, post- treatment: 4 weeks, follow-up: 3 mos.	n=13/35, (CBT=7/18, WLC=6/17)
Stinson et al 2010, Canada	Juvenile idiopathic arthritis	N= 46, 32F (70%), 12-18 yrs.; (M = 14.6, SD = 1.5)	Pilot, ITT	Clinic (2 mos.)	Internet based CBT (n = 22), + email contact, 12 weeks, 12 online modules, over avg. 14.7 wks. (360 mins)	Attention control (n=24), weekly phone call	12 weeks (360 mins)	Baseline, post- treatment: 12 wks., 12 mos.	n=6/46, (CBT=2/22, WLC=4/24)

CBT: cognitive behaviour therapy; WLC: waitlist control; ITT: intent to treat; NR: not reported; N: total sample size; n: group sample size

Table 3.2

Characteristics of included studies continued

Study	Pain condition	Participants N (% female), age range (mean / SD)	Design and analysis	Recruitment (period)	Intervention (N)	Control (N)	Duration of treatment (mins)	Timing of outcome assessment	Withdrawal (intervention, control)
Trautmann & Kroner-Herwig 2010, Germany	Headache	N= 65, 36F (55%), 9-16 yrs., (M = 12.6, SD= 2.1)	RCT	Community, 1.2 yr. recruitment period	Internet based CBT (n = 24) & applied relaxation (n = 22) (AR), + email contact, 8 sessions / 8 weeks (NR mins)	Internet educational module (n = 19), + email contact	8 weeks (NR)	Baseline, post-treatment: 6 weeks, follow-up: 6 mos.	n=25/45, (CBT=13/24, AR= 3/22), EDU: (9/19)

CBT: cognitive behaviour therapy; WLC: waitlist control; ITT: intent to treat; NR: not reported; N: total sample size; n: group sample size

Table 3.3

Summary of findings for comparison of clinical outcomes

Study	Outcomes (Measures)	Post-treatment effect on clinical outcome	Follow-up effect on clinical outcome
Connelly et al 2006,	Clinically significant improvement in total pain at post-treatment and follow-up (Headache Index).	Yes; I = 7/14; C = 0/17	Yes; I = 7/14; C = 7/17
	Headache index (NRS)	Improved [#]	No effect
	Headache frequency (NRS)	Improved [#]	No effect
	Headache severity (NRS)	Improved [#]	No effect
	Headache duration (NRS)	Improved [#]	No effect
	Headache related disability (The Pediatric Migraine Disability Assessment)	No effect	No effect
Cottrell et al., 2007,	Clinically significant reduction in headache pain severity.	Yes; I = 12/15; C = NR	Yes; I = 11/14; C = NR
	Headache diary;	Improved*	Improved ^a
	Quality of Life: Migraine Specific Quality of Life Questionnaire-Adolescent;	Improved*	
Hicks et al., 2006,	Clinically significant reduction in headache pain severity (Total pain).	Yes; I = 15/21; C = 3/16	Yes; I = 3/18; C = 2/14
	Pediatric Quality of Life Inventory (PEDsQL).	No effect	No effect

No effect: no statistically significant effect; *denotes a significant change from T1 to T2 in a group; #denotes a significant difference between groups at T2; ^adenotes a significant change from T1 to T3 in Internet group; NR: not reported; VAS: visual analogue scale; NRS: numerical rating scale;

Table 3.3

Summary of findings for comparison of clinical outcomes continued

Study	Outcomes (Measures)	Post-treatment effect on clinical outcome	Follow-up effect on clinical outcome
Law 2015,	Clinically significant reduction in headache pain severity.	Yes; I = 12/31; C = 7/28 (both groups)	Yes; I = 19/31; C = 10/28 (both groups)
	Headache frequency (Total headache free days)	Improved* (both groups)	Improved ^a (both groups)
	Pain intensity (Pain diary)	Improved* (both groups)	Improved ^a (both groups)
	Activity limitations (Child Activity Limitations Interview, CALI)	Improved* (both groups)	Improved ^a (both groups)
	Depression (Children's Depression Inventory, CDI);	Improved* (both groups)	Improved ^a (both groups)
	Anxiety (Revised Children's Manifest Anxiety Scale-Second Edition, RCMAS-2)	No effect	No effect
	Protective response (Protect subscale Adult Responses to Children's Symptoms, ARCS).	Improved* (both groups)	Improved* (both groups)
McGrath 1992,	Clinically significant reduction in headache pain severity.	Yes; I = 10/23; C = 16/24	NR

No effect: no statistically significant effect; *denotes a significant change from T1 to T2 in a group; #denotes a significant difference between groups at T2; ^adenotes a significant change from T1 to T3 in Internet group; NR: not reported; VAS: visual analogue scale; NRS: numerical rating scale;

Table 3.3

Summary of findings for comparison of clinical outcomes continued

Study	Outcomes (Measures)	Post-treatment effect on clinical outcome	Follow-up effect on clinical outcome
Palermo et al 2009,	Clinically significant reduction in headache pain severity (Total pain).	Yes; I = 10/23; C = 3/21	NR
	Pain interference (Child Activity Limitations Interview; CALI)	Improved*#	Improved ^a
	Pain intensity (Pain diary, NRS)	Improved#	No effect
	Depression: (Major depressive disorder subscale of the Revised Child Anxiety and Depression Scale; RCADS)	No effect	Improved ^a
	Treatment Satisfaction (Treatment Evaluation Inventory-Short Form)	No effect	NR
	Parental response (Protect subscale -Adult Responses to Children's Symptoms, ARCS).	No effect	NR
Palermo et al., 2016	Pain diary (NRS)	No effect	No effect
	Pain interference (Child Activity Limitations Interview; CALI)	No effect	Improved# ^a
	Depression Bath Adolescent Pain Questionnaire (BAPQ)-Depression subscale	Improved#	No effect
	Anxiety Bath Adolescent Pain Questionnaire (BAPQ)-Anxiety subscale	Improved#	No effect
	Treatment Satisfaction (Treatment Evaluation Inventory-Short Form)	Improved#	Improved#
	Parental response (Protect subscale -Adult Responses to Children's Symptoms, ARCS).	Improved#	Improved#

No effect: no statistically significant effect; *denotes a significant change from T1 to T2 in a group; # denotes a significant difference between groups at T2; ^adenotes a significant change from T1 to T3 in Internet group; NR: not reported; VAS: visual analogue scale; NRS: numerical rating scale;

Table 3.3

Summary of findings for comparison of clinical outcomes continued

Study	Outcomes (Measures)	Post-treatment effect on clinical outcome	Follow-up effect on clinical outcome
Rapoff 2014	Headache pain severity (Headache diary; VAS)	Improved* Yes; I = 7/18; C = 6/17	Yes; I = 7/11; C = 7/11
	The Pediatric Migraine Disability Assessment (PedsMIDAS)	No effect	No effect
	Pediatric Quality of Life Inventory (PEDsQL)	No effect	No effect
Stinson et al 2010,	Juvenile Arthritis Quality of Life Questionnaire (JAQQ),	No effect	NR
	Recalled Pain Inventory (RPI)	Improved [#]	NR
	Children's Arthritis Self-Efficacy (CASE)	No effect	NR
	Treatment acceptability, Satisfaction and Expectation (NRS)		NR
Trautmann & Kroner-Herwig 2010,	Clinically significant reduction in headache pain severity (Headache diary)	Improved* I = 16/35; C = 2/16	NR
	Pain intensity (Pain diary (NRS);	No effect	No effect
	Pain Catastrophizing Scale for Children (PCS-C);	Improved* (all groups)	Improved ^a (all groups)
	Children's Depression Inventory (CDI);	No effect	No effect
	Health-related quality of life (KINDL-R)	No effect	No effect
	Treatment Evaluation (NRS)	No effect	No effect

No effect: no statistically significant effect; *denotes a significant change from T1 to T2 in a group; # denotes a significant difference between groups at T2; ^adenotes a significant change from T1 to T3 in Internet group; NR: not reported; VAS: visual analogue scale; NRS: numerical rating scale;

3.6.4 Excluded Studies

At the point of full-text screening, five studies were excluded from this review. Two studies were excluded due to insufficient sample size, as they included fewer than 10 participants in each arm of the trial at post-treatment (Merlijn et al., 2005; Trautmann & Kröner-Herwig, 2008). One study was excluded due to insufficient therapeutic content in the intervention arm of the trial (Stinson et al., 2016). One study was excluded because it is an open trial (Bonnert et al., 2014) and the study by Long and Palermo (2009) was excluded on the basis that it was a usability study that is linked to a trial already included in the review (Palermo et al., 2009).

3.6.5 Assessment of risk of bias

A summary of the risk of bias ratings by domain and by study for all 10 trials are shown in Figures 3.2 and 3.3. Detailed information on risk of bias for all 10 trials is reported in Appendix 9: Characteristics of included studies.

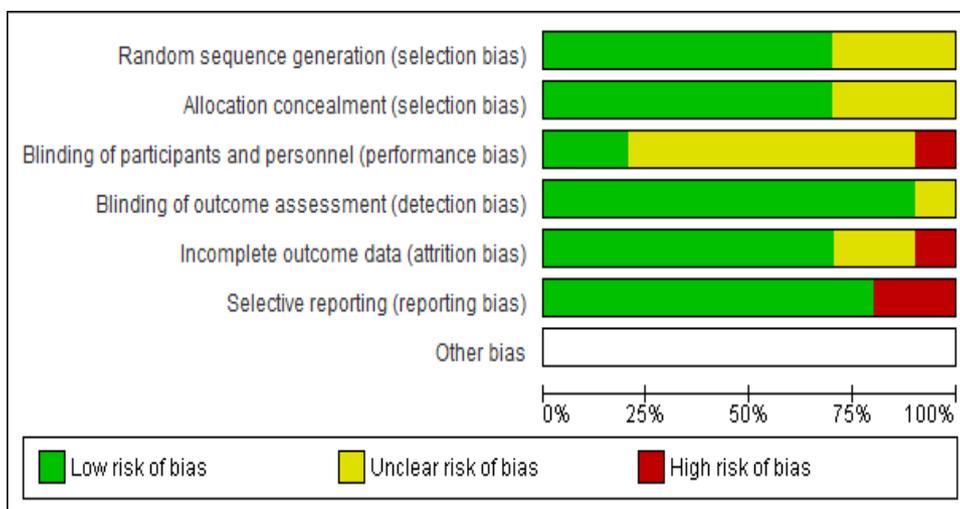


Figure 3.2 Summary of risk of bias ratings by domain

	Random sequence generation (selection bias)	Allocation concealment (selection bias)	Blinding of participants and personnel (performance bias)	Blinding of outcome assessment (detection bias)	Incomplete outcome data (attrition bias)	Selective reporting (reporting bias)	Other bias
Connelly 2006	+	+	+	+	+	+	
Cottrell 2007	?	?	?	?	?	-	
Hicks 2006	+	+	?	+	?	+	
Law 2015	+	+	-	+	+	+	
McGrath 1992	?	?	?	+	+	-	
Palermo 2009	+	+	?	+	+	+	
Palermo 2016	+	+	+	+	+	+	
Rapoff 2014	?	?	?	+	-	+	
Stinson 2010	+	+	?	+	+	+	
Trautmann 2010	+	+	?	+	+	+	

Figure 3.3 Summary of risk of bias ratings by study

3.6.6 Quality of evidence

Six trials were rated as unclear risk of bias across several domains reducing the overall quality of evidence. Most studies did not provide enough information to judge whether blinding procedures for participants or personnel were adequate. Of the ten studies examined, only three gave an adequate description of blinding of participants and personnel (Connelly et al., 2006; Law et al., 2015; Palermo et al., 2016) and only two trials were rated as low risk of bias across all domains (Connelly et al., 2006; Palermo et al., 2016). Most included studies report intention to treat analyses and adequate report rates of attrition and were graded as low risk of bias as a result. However, all but one study (Stinson et al., 2010a) failed to fully report reasons for attrition or difference in the profile of participants withdrawing from the trial. Other sources of potential bias were considered.

To the best of our knowledge, none of the included trials were linked to a published protocol which raises concern regarding selective reporting bias. Also, for all studies the outcome measures used were self-report and results may have been influenced by bias. GRADE was used to assess the quality of evidence. None of the included outcomes were rated as high quality. Four outcomes were rated as moderate in quality at post-treatment or follow-up and five outcomes were rated as low quality at post-treatment or follow-up. Four outcomes were rated as very low quality at post-treatment or follow-up. A summary of findings is presented separately for headache and mixed pain conditions in Tables 3.4 and 3.5.

Table 3.4

Summary of findings – headache conditions

ICT-based psychological therapies compared to Waitlist or active control conditions for chronic pain in children and adolescents						
Patient or population: patients with chronic pain in children and adolescents						
Settings: Information and communication technology						
Intervention: ICT-based psychological therapies						
Comparison: Waitlist or active control conditions						
Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)	Comments
	Assumed risk	Corresponding risk				
	Waitlist or active control conditions	ICT-based psychological therapies				
Reduction in headache severity (post-treatment) Clinically significant reduction in headache pain defined as >50% reduction Follow-up: 4-10 weeks	Study population		OR 3.44 (1.73 to 6.81)	328 (7 studies)	⊕⊕⊕⊖ moderate ¹	
	208 per 1000	475 per 1000 (312 to 641)				
	Moderate					
	180 per 1000	430 per 1000 (275 to 599)				

Reduction in headache severity (follow-up) Clinically significant reduction in headache pain defined as >50% reduction Follow-up: 3 months	Study population		OR 2.44 (0.92 to 6.45)	168 (4 studies)	⊕⊕⊖⊖ low ^{2,3,4}	
	321 per 1000	536 per 1000 (303 to 753)				
	Moderate					
	334 per 1000	550 per 1000 (316 to 764)				
Disability (post-treatment) Investigators measured disability using different instruments. Low scores indicate lower disability. Follow-up: 4-10 weeks	The mean disability (post-treatment) in the intervention groups was 0.15 standard deviations lower (0.43 lower to 0.13 higher)			472 (5 studies)	⊕⊕⊕⊖ moderate ⁴	SMD -0.15 (-0.43 to 0.13)
Disability (follow-up) Investigators measured disability using different instruments. Low scores indicate lower disability.	The mean disability (follow-up) in the intervention groups was 0.19 standard deviations lower (0.47 lower to 0.09 higher)			387 (3 studies)	⊕⊕⊖⊖ low ^{1,2,4}	SMD -0.19 (-0.47 to 0.09)
Depression (post-treatment) Investigators measured	The mean depression (post-treatment) in the intervention groups was			455 (4 studies)	⊕⊕⊕⊖ moderate ⁴	SMD 0.03 (-0.16 to 0.21)

depression using different instruments. Low scores indicate lower levels of depression.	0.03 standard deviations higher (0.16 lower to 0.21 higher)			
Depression (follow-up) Investigators measured depression using different instruments. Low scores indicate lower levels of depression.	The mean depression (follow-up) in the intervention groups was 0.04 standard deviations higher (0.16 lower to 0.24 higher)	387 (3 studies)	⊕⊕⊕⊖ moderate ^{1,4}	SMD 0.04 (-0.16 to 0.24)
Quality of life (post-treatment) Investigators measured quality of life using different instruments. Low scores indicate higher quality of life.	The mean quality of life (post-treatment) in the intervention groups was 0.05 standard deviations higher (0.29 lower to 0.4 higher)	136 (3 studies)	⊕⊖⊖⊖ very low ^{1,2,4}	SMD 0.1 (-0.25 to 0.45)
Quality of life (follow-up) Investigators measured quality of life using different instruments. Low scores indicate higher quality of life.	The mean quality of life (follow-up) in the intervention groups was 0.01 standard deviations higher (0.36 lower to 0.38 higher)	123 (3 studies)	⊕⊖⊖⊖ very low ^{1,2,4}	SMD -0.08 (-0.45 to 0.3)

Treatment satisfaction (post-treatment) Investigators measured disability using different instruments. Low scores indicate greater satisfaction Follow-up: 8-10 weeks	The mean treatment satisfaction (post-treatment) in the intervention groups was 0.39 standard deviations lower (0.71 to 0.08 lower)	415 (3 studies)	⊕⊕⊕⊖ moderate ^{4,5}
Treatment satisfaction (follow-up) Investigators measured disability using different instruments. Low scores indicate greater satisfaction Follow-up: 3-6 months	The mean treatment satisfaction (follow-up) in the intervention groups was 0.35 standard deviations lower (0.56 to 0.14 lower)	352 (2 studies)	⊕⊕⊕⊖ moderate ^{1,4}

*The basis for the **assumed risk** (e.g. the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% confidence interval) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **OR:** Odds ratio;

GRADE Working Group grades of evidence

High quality: Further research is very unlikely to change our confidence in the estimate of effect.

Moderate quality: Further research is likely to have an important impact on our confidence in the estimate of effect and may change the estimate.

Low quality: Further research is very likely to have an important impact on our confidence in the estimate of effect and is likely to change the

estimate.

Very low quality: We are very uncertain about the estimate.

¹ Number of events does not meet the optimal information size as per the 'rules of thumb' (e.g. >400 events).

² Risk of bias unclear or high in several domains

³ I squared = 46% (moderate to substantial) heterogeneity, Chi squared = $p > 0.05$

⁴ Confidence intervals include the possibility of a small or no effect.

⁵ I squared = 49% (moderate to substantial) heterogeneity, Chi squared = $p > 0.05$

Table 3.5

Summary of findings – mixed pain conditions

ICT-based psychological therapies for mixed conditions: treatment versus control (post-treatment) for chronic pain in children and adolescents						
Patient or population: patients with chronic pain in children and adolescents						
Settings:						
Intervention: ICT-based psychological therapies for mixed conditions: treatment versus control (post-treatment)						
Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)	Comments
	Assumed risk	Corresponding risk				
	Control	ICT-based psychological therapies for mixed conditions: treatment versus control (post-treatment)				
Pain intensity (post-treatment) Investigators measured intensity using different instruments. Low scores indicate lower intensity.		The mean pain intensity (post-treatment) in the intervention groups was 0.37 standard deviations lower (0.86 lower to 0.11 higher)		410 (4 studies)	⊕⊕⊖⊖ low ^{1,2}	SMD -0.37 (-0.86 to 0.11)
Pain Intensity (follow-up) Investigators measured intensity		The mean pain intensity (follow-up) in the intervention groups was		316 (2 studies)	⊕⊖⊖⊖ very low ^{2,3,4}	SMD -0.45 (-1.68 to 0.79)

using different instruments. Low scores indicate lower intensity. Follow-up: 3-6 months	0.45 standard deviations lower (1.68 lower to 0.79 higher)			
Disability (post-treatment) Investigators measured disability using different instruments. Low scores indicate lower disability.	The mean disability (post-treatment) in the intervention groups was 0.33 standard deviations lower (1.09 lower to 0.42 higher)	317 (2 studies)	⊕⊕⊕⊕ very low ^{2,4,5}	SMD -0.33 (-1.09 to 0.42)
Depression (post-treatment) Investigators measured disability using different instruments. Low scores indicate lower disability.	The mean depression (post-treatment) in the intervention groups was 0.04 standard deviations higher (0.18 lower to 0.26 higher)	317 (2 studies)	⊕⊕⊕⊕ low ^{2,4}	SMD 0.04 (-0.18 to 0.26)
Quality of life (post-treatment) Investigators measured quality of life using different instruments. Low scores indicate lower quality of life. Follow-up: 4-12 weeks	The mean quality of life (post-treatment) in the intervention groups was 0 standard deviations higher (0.46 lower to 0.46 higher)	93 (2 studies)	⊕⊕⊕⊕ low ^{2,4}	SMD 0 (-0.46 to 0.46)

*The basis for the **assumed risk** (e.g. the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% confidence interval) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval;

GRADE Working Group grades of evidence

High quality: Further research is very unlikely to change our confidence in the estimate of effect.

Moderate quality: Further research is likely to have an important impact on our confidence in the estimate of effect and may change the estimate.

Low quality: Further research is very likely to have an important impact on our confidence in the estimate of effect and is likely to change the estimate.

Very low quality: We are very uncertain about the estimate.

¹ I squared = 76% (substantial-considerable) heterogeneity, Chi squared = $p < 0.001$

² Confidence intervals include the possibility of a small or no effect

³ I squared = 93% (considerable) heterogeneity Chi squared = $p < 0.001$

⁴ Number of events does not meet the optimal information size as per the 'rules of thumb' (e.g. >400 events).

⁵ I squared = 83% (considerable) heterogeneity Chi squared = $p < 0.05$

3.6.7 Meta-analytic strategy

Table 3.6. shows the overall results of the meta-analysis for all outcomes at post-treatment and follow-up. Forest plots for each of the primary analyses of the impact of interventions on end-point outcomes are presented below. Meta-analyses were conducted separately for headache and mixed pain conditions.

3.6.8 Meta-analysis - Headache Pain Conditions

3.6.9 Effects on headache severity

Treatment efficacy data (defined as >50% reduction in headache severity) was available for 7 trials (n = 328 participants) at post-treatment (varying from 4 to 10 weeks after baseline). Odds ratios varied from 1.17 to 35.00 with an overall mean of 3.51 (95% CI [1.79, 6.86], $z = 3.67$, $p = .0002$), number need to treat to benefit (NNTB) 3.91; Figure 3.4). The rate of reduction in headache severity among the treatment group (n = 166) was 50% compared to 22.5% for controls (n = 138). Heterogeneity between the studies was low to moderate and not significant ($I^2 = 31\%$, $p = 0.19$). The quality rating based on GRADE guidelines was moderate for this outcome, indicating that we can be moderately confident in this estimate of effect. Peter's regression test showed no evidence of asymmetry in the ES funnel plot at post-treatment ($p=0.54$).

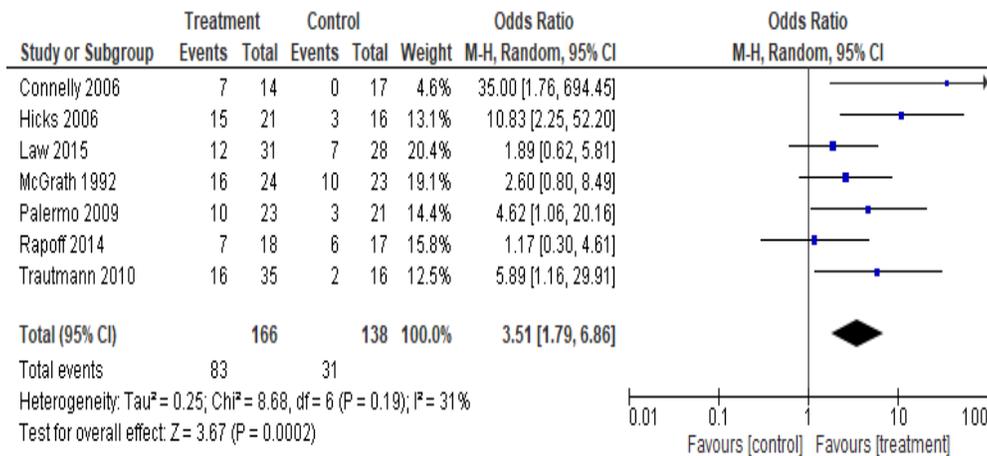


Figure 3.4 Reduction in headache severity at post-treatment, treatment versus control ('as treated')

Treatment efficacy data was available for four trials (N = 134 participants) at follow-up (approximately 3 months). Beneficial effects were not maintained at follow-up. The rate of reduction in headache severity among treatment participants (n = 46) was 66% compared to 41% for controls (n = 26). Odds ratios varied from 1.00 to 15.60 with an overall mean of 2.67 (95% CI [0.95, 7.50], $z = 1.87$, $p = 0.06$). Heterogeneity between the studies was moderate but not significant ($I^2 = 47%$, $p = 0.13$; Figure 3.5). Peter's regression test showed no evidence of asymmetry in the ES funnel plot at follow-up ($p = 1.00$). The quality rating for this outcome at follow-up was judged to be low meaning further research is very likely to have an important impact on our confidence in the estimate of effect and very likely to change the estimate. At post-treatment and follow-up assessment the effects varied widely from trial to trial. The largest effect sizes were found in the trial conducted by Connelly et al. (2006) at post-treatment ($d = 35.00$) and the trial by Hicks et al. (2006) at follow-up ($d = 15.60$). The smallest effects were found in the trial conducted by Rapoff et al. (2014) at post-treatment ($d = 1.17$) and follow-up ($d = 1.00$).

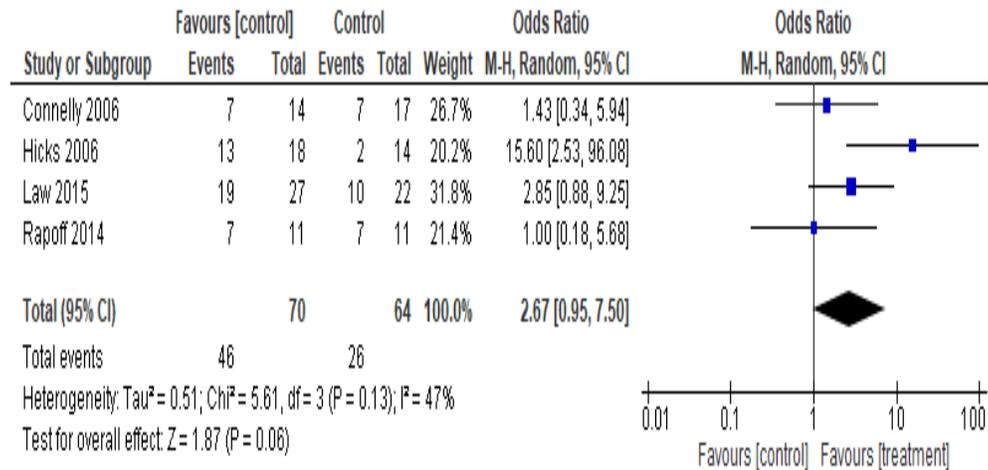


Figure 3.5 Reduction in headache severity at follow-up, treatment versus control ('as treated')

3.6.10 Effects on headache related pain interference

Five trials were included in analyses comparing ICT-delivered therapies to controls (Connolly et al., 2006, Palermo et al., 2009, Rapoff et al., 2014, Law et al., 2015, Palermo et al., 2016) on measures of headache pain interference at post-treatment (4-10 weeks). The sample included data on 472 participants. The ICT-delivered therapies had no effect, standardised mean difference (SMD) - 0.15 (95% CI [-0.43, 0.13], $z = 1.03$, $p = 0.30$, Figure 3.6). Egger's regression test showed no evidence of asymmetry in the ES funnel plot at post-treatment ($p = 0.35$).

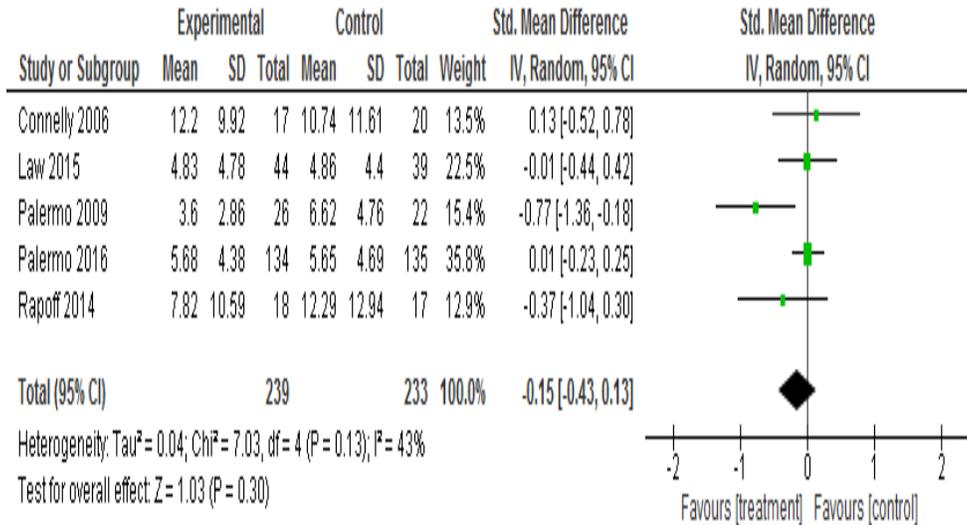


Figure 3.6 Headache pain conditions, disability at post-treatment, treatment versus control.

Of the five studies examined, only three (Rapoff et al., 2014, Law et al., 2015 and Palermo et al., 2016) provided follow-up data. At follow-up (varying from 3 to 6 months), the sample included data on 387 participants. The ICT-delivered therapies had no effect, standardised mean difference (SMD) -0.19 (95% CI [-0.47, 0.09], $z = 1.33$, $p = 0.18$, Figure 3.7). The largest effect sizes were found in the trial conducted by Palermo et al. (2009) at post-treatment ($d = -0.77$) and the trial by Rapoff et al. (2014) at follow-up ($d = -0.71$). The trial conducted by Connelly et al. (2006) showed negative effects ($d = 0.13$) at post-treatment. Egger's regression test showed no evidence of asymmetry in the ES funnel plot at follow-up ($p = 0.58$). The quality rating based on GRADE guidelines was moderate for this outcome at post-treatment indicating we can be moderately confident in this estimate of effect. However, quality was rated as low at follow-up meaning further research is very likely to have an important impact on our confidence in the estimate of effect and is likely to change the estimate of effect.

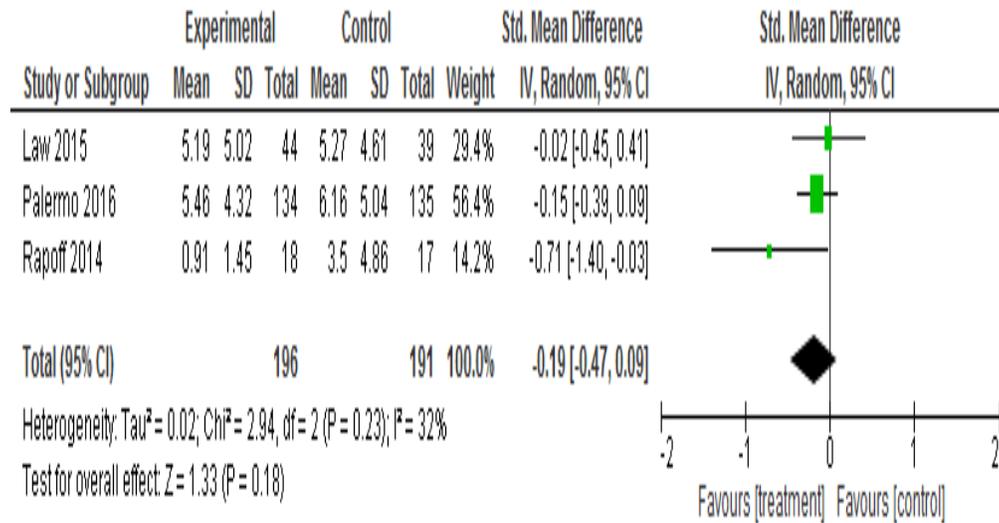


Figure 3.7 Headache pain conditions, pain interference at follow-up, treatment versus control.

3.6.11 Effects on depression

Four trials were included in analyses comparing ICT-delivered therapy to controls (Palermo et al., 2009; Trautmann & Kröner-Herwig, 2010; Law et al., 2015; Palermo et al., 2016) on measures of depression. The sample included data on 455 participants at post-treatment (approximately 8-10 weeks). The ICT-delivered therapies had no effect, standardised mean difference (SMD) 0.03 (95% CI [-0.16, 0.21], $z = 0.29$, $p = 0.77$, Figure 3.8). Egger's regression test showed no evidence of asymmetry in the ES funnel plot ($p=0.64$).

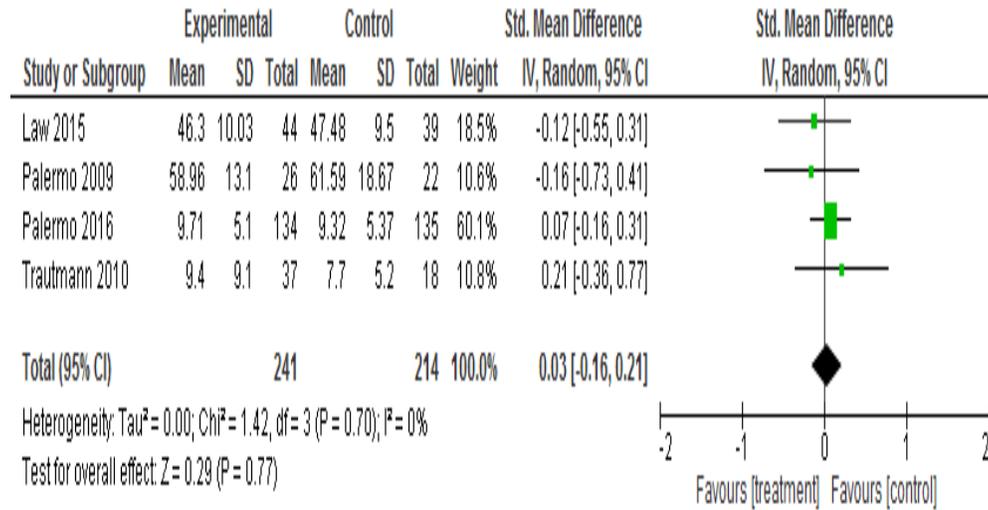


Figure 3.8 Headache pain conditions, depression at post-treatment, treatment versus control.

Three trials were included in follow-up analyses. At follow-up (varying from 3 to 6 months) ICT-delivered therapies had no effect, (SMD) 0.04 (95% CI [-0.16, 0.24], $z = 0.40$, $p = 0.69$, Figure 3.9). The largest effect sizes were found in the trial conducted by Trautmann and Kröner-Herwig (2010) at post-treatment ($d = 0.21$) in favour of the control condition. Egger's regression test showed no evidence of asymmetry in the ES funnel plot at follow-up ($p=0.47$). This outcome was rated as moderate quality at post-treatment and follow-up and further research is likely to have an important impact on our confidence in the estimate of effect and may change the estimate.

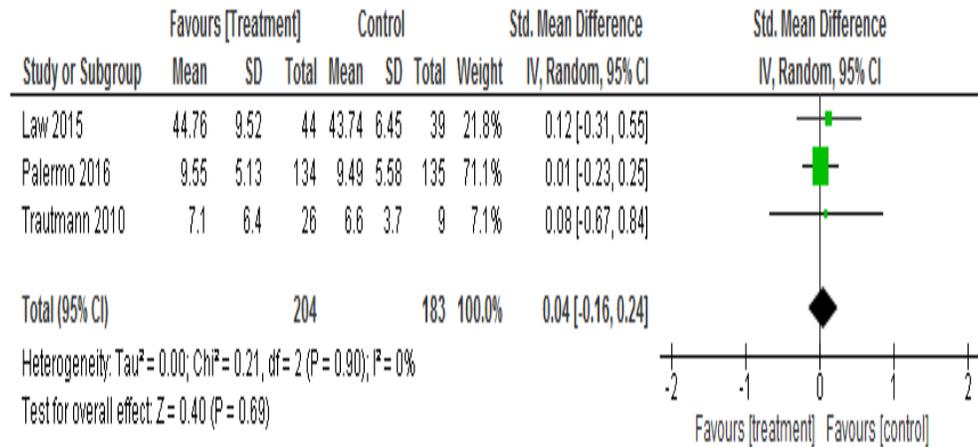


Figure 3.9 Headache pain conditions, depression at follow-up, treatment versus control.

3.6.12 Effects on anxiety

Anxiety was reported in only one study investigating the effects of treatment on headache pain conditions (Palermo et al., 2016), therefore quantitative and quality analyses were not conducted.

3.6.13 Effects on quality of life

Three studies were included in analyses comparing ICT-delivered therapy to controls (Hick et al., 2006; Rapoff et al., 2014; Trautmann & Kröner-Herwig, 2010) on measures of quality of life. The sample included data on 136 participants. The ICT-delivered therapies had no effect, standardised mean difference (SMD) 0.05 (95% CI [-0.29, 0.40], $z = 0.29$, $p = 0.77$, Figure 3.10). Egger's regression test showed no evidence of asymmetry in the ES funnel plot at post-treatment ($p=0.20$).

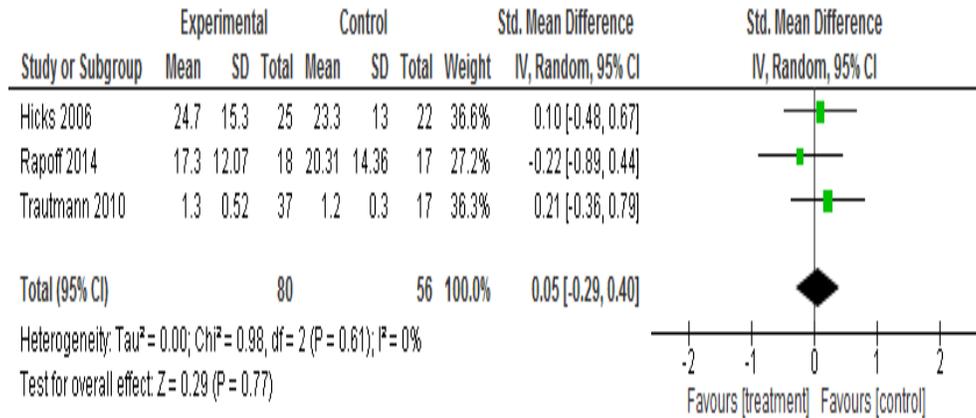


Figure 3.10 Headache pain conditions, quality of life at post-treatment, treatment versus control.

Follow-up data suggest no effects of ICT- delivered therapy over time, (SMD) 0.01 (95% CI [-0.36, 0.38], $z = 0.06$, $p = 0.95$, Figure 3.11). Egger's regression test showed no evidence of asymmetry in the ES funnel plot at follow-up ($p = 0.23$). This outcome was rated as very low quality at post-treatment and follow-up meaning we are very uncertain about the estimate.

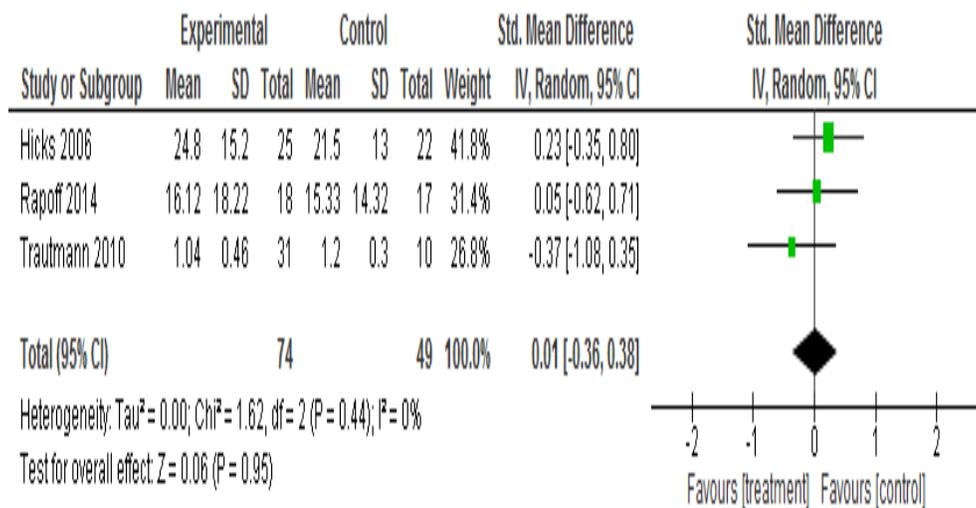


Figure 3.11 Headache pain conditions, quality of life at follow-up, treatment versus control.

3.6.14 Effect on treatment satisfaction

Three trials (Trautmann & Kröner-Herwig, 2010; Law et al., 2015; Palermo et al., 2016) investigated treatment satisfaction in response to ICT-delivered treatment compared with control conditions among children with headache pain. Only three trials were included in analyses due to the heterogeneity of the measures used and a lack of comparative data resulting from the use of passive control conditions. The ICT-delivered therapies had a significant effect in favour of the treatment group, standardised mean difference (SMD) -0.39 (95% CI [-0.71, -0.08], $z = 2.42$, $p = 0.02$, Figure 3.12).

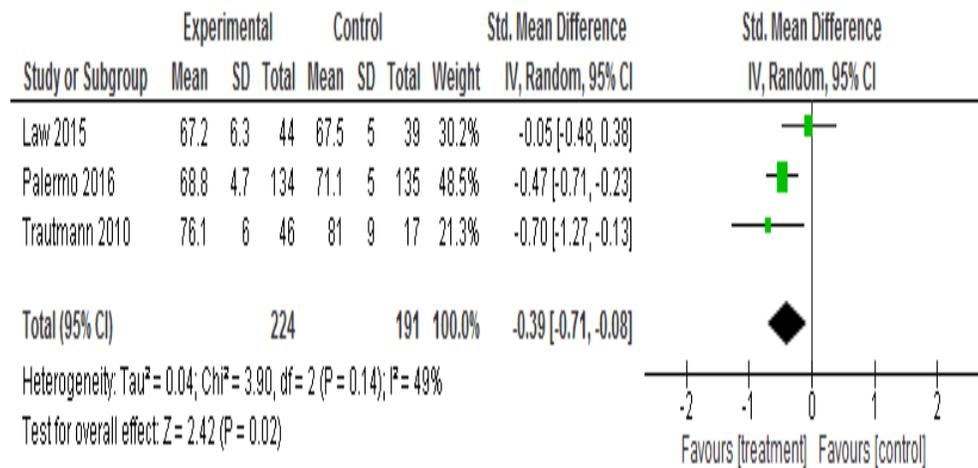


Figure 3.12 Headache pain conditions, treatment satisfaction at post-treatment, treatment versus control.

Only two studies requested comparative data from control groups at follow-up and were included in analyses (Law et al., 2015; Palermo et al., 2016). Follow-up data suggest treatment satisfaction in favour of the treatment group was maintained over time, standardised mean difference (SMD) -0.35 (95% CI [-0.56, -0.14], $z = 3.22$, $p = .001$, Figure 3.13). The quality rating based on GRADE guidelines was rated as moderate for this outcome at post-treatment and follow-up, indicating we can be moderately confident in this estimate of effect.

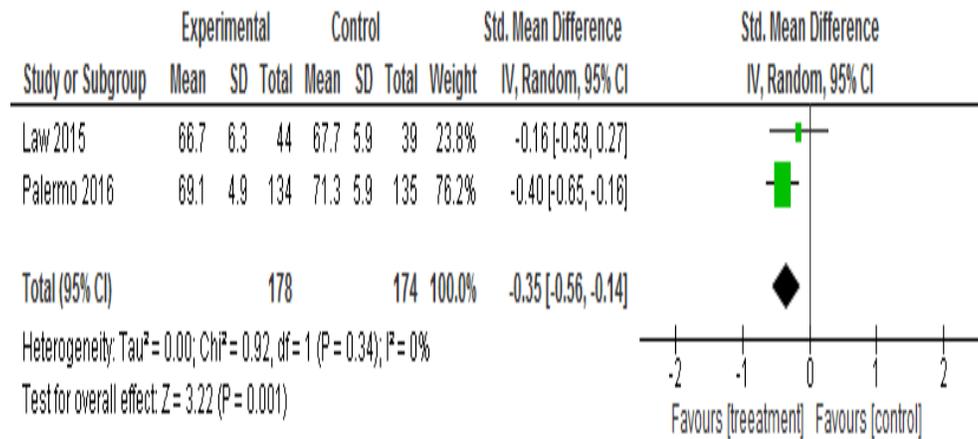


Figure 3.13 Headache pain conditions, treatment satisfaction at follow-up, treatment versus control.

3.6.15 Meta-analysis – Mixed Pain Conditions Pain intensity

Four studies were included in analyses comparing ICT-delivered treatments to controls (Hicks et al., 2006; Palermo et al., 2009; Stinson et al., 2010a; Palermo et al., 2016) on measures of pain intensity. The sample included data on 410 participants. The ICT-delivered therapies had no effect, standardised mean difference (SMD) -0.37 (95% CI [-0.86, 0.11], $z = 1.50$, $p = 0.13$, Figure 3.14).

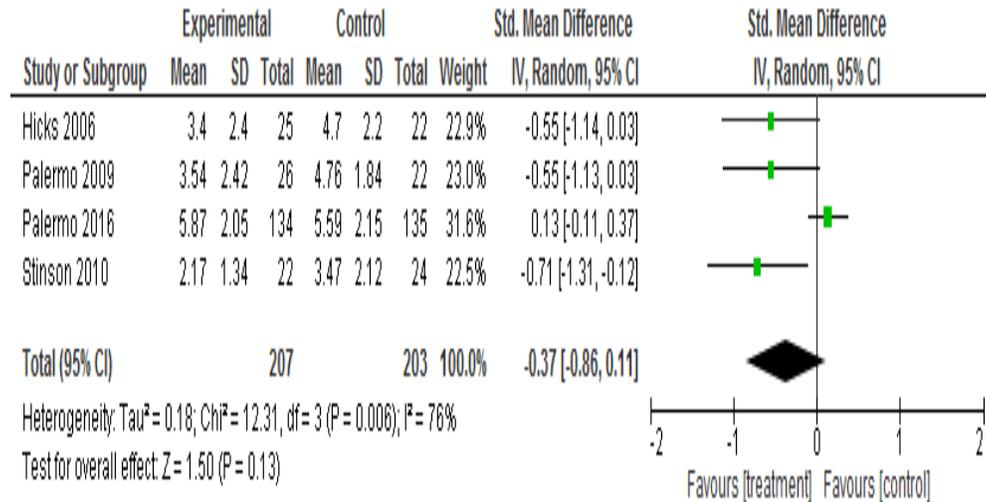


Figure 3.14 Mixed pain conditions, pain intensity at post-treatment, treatment versus control.

Egger's regression test showed borderline evidence of asymmetry in the ES funnel plot at post-treatment ($p = 0.05$). The quality rating for this outcome was low post-treatment meaning further research is very likely to have an important impact on our confidence in the estimate of effect and may change the estimate. Follow-up data suggest no effects of ICT-delivered therapy over time, (SMD) -0.45 (95% CI [-1.68, 0.79], $z = 0.71$, $p = 0.48$, Figure 3.15). Egger's regression analysis could not be performed due to a lack of data at follow-up. This outcome was rated as very low quality at post-follow-up meaning we are very uncertain about the estimate.

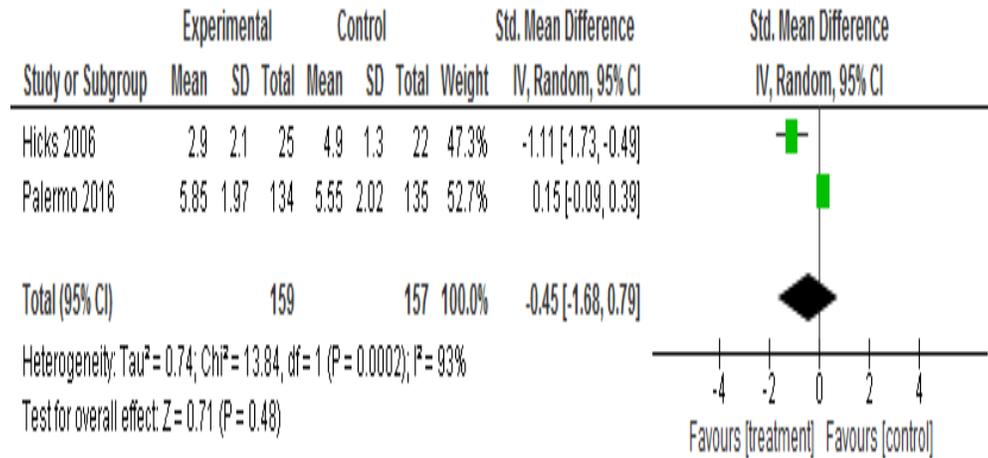


Figure 3.15 Mixed pain conditions, pain intensity at follow-up, treatment versus control.

3.6.16 Pain interference

Two studies were included in analyses comparing ICT-delivered treatments to controls (Palermo et al., 2009; Palermo et al., 2016) on measures of pain interference. The sample included data on 317 participants. The ICT-delivered therapies had no effect, standardised mean difference (SMD) -0.33 (95% CI [-1.09, 0.42], $z = 0.86$, $p = 0.39$, Figure 3.16). Egger's regression analysis could not be performed due to a lack of data at post-treatment and follow-up. This outcome was rated as very low quality at post-treatment very low quality at post-treatment and follow-up meaning we are very uncertain about the estimate. Of the two studies only one reported follow-up data therefore follow-up analyses were not conducted.

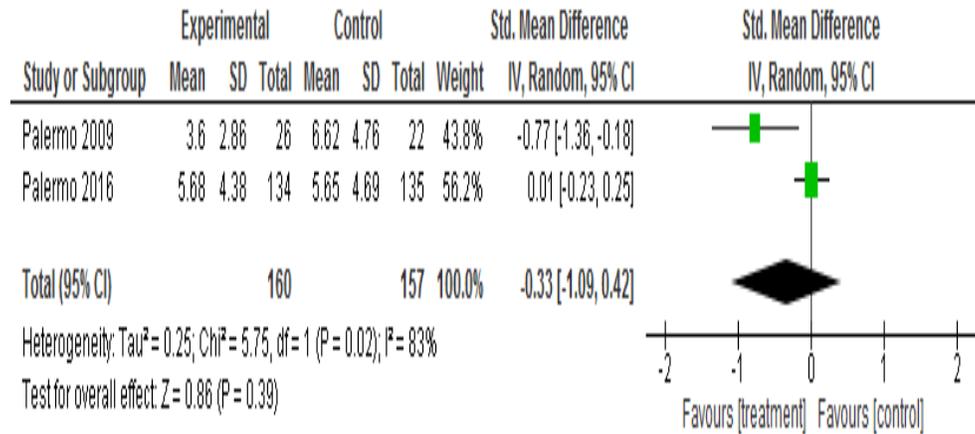


Figure 3.16 Mixed pain conditions, disability at post-treatment, treatment versus control.

3.6.17 Depression

Two studies were included in analyses comparing ICT-delivered treatments to controls (Palermo et al., 2009; Palermo et al., 2016) on measures of depression. The sample included data on 317 participants. The ICT-delivered therapies had no effect, standardised mean difference (SMD) 0.04 (95% CI [-0.18, 0.26], $z = 0.34$, $p = 0.73$, Figure 3.17).

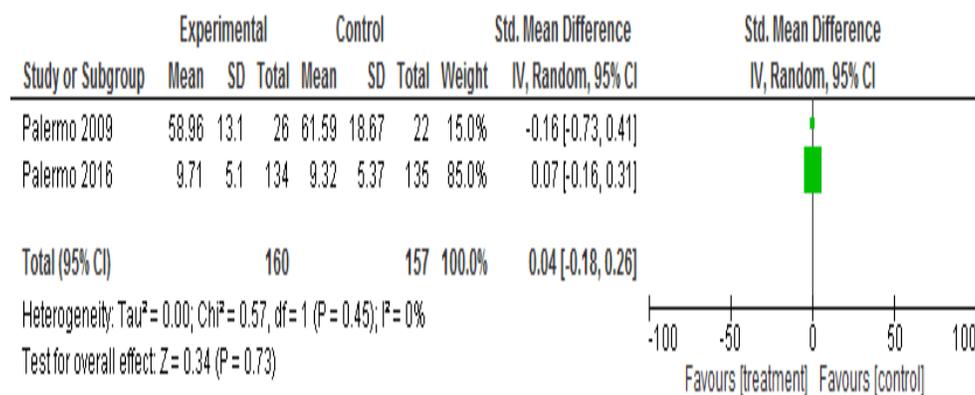


Figure 3.17 Mixed pain conditions, depression at post-treatment, treatment versus control.

Egger's regression analysis could not be performed due to a lack of data at post-treatment and follow-up. This outcome was rated as low quality at post-treatment and further research is very likely to have an important impact on our confidence in the estimate of effect and is likely to change the estimate of effect. Of the two studies only one (Palermo et al., 2016) reported follow-up data therefore follow-up analyses were not conducted.

3.6.18 Anxiety

Only one study reported the effects of ICT-delivered treatment on levels of anxiety for children with mixed pain conditions (Palermo et al., 2016), therefore quantitative and quality analyses were not conducted.

3.6.19 Quality of life

Two studies investigated the effects of ICT-delivered treatment on quality of life for children with mixed pain. The ICT-delivered therapies had no effect, standardised mean difference (SMD) -0.07 (95% CI [-0.48, 0.34], $z = 0.34$, $p = 0.74$, Figure 3.18). Egger's regression analysis could not be performed due to a lack of data at post-treatment and follow-up. This outcome was rated as low quality at post-treatment meaning we have low confidence in this estimate of effect and further research is likely to change the estimate of effect. Only one study (Hicks, 2006) reported follow-up data therefore follow-up analyses were not conducted.

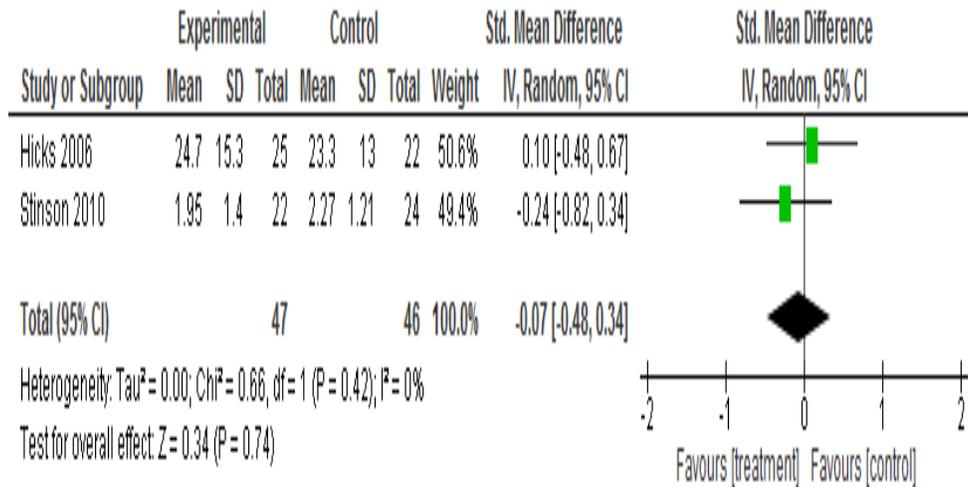


Figure 3.18 Mixed pain conditions, quality of life at post-treatment, treatment versus control.

3.6.20 Parental Protectiveness: ICT treatments versus controls

Two studies (Palermo et al., 2009; Palermo et al., 2016) investigated the effects of ICT-delivered treatment on parental protective behaviour for children with headache and mixed pain conditions. However, only one study provided child reports of parental protective behaviour (Palermo et al., 2009) therefore quantitative and quality analyses were not conducted.

Table 3.6

Effects of the interventions for each outcome at post-treatment and follow-up

Outcomes	Assessment point	No. of trials	Sample size	<i>d</i> (95% CI)	Z	Q ^b	I ²
Headache Pain Conditions							
Reduction in headache pain	Post-treatment	7	304	3.51 (1.79, 6.86)	3.67***	8.68	31%
	Follow-up	4	144	2.67 (0.98, 7.30)	1.92†	5.62	47%
Disability	Post-treatment	5	472	-0.15 (-0.43, 0.13)	1.03	7.03	43%
	Follow-up	3	387	-0.19 (-0.47, 0.09)	1.33	2.94	32%
Depression	Post-treatment	4	455	0.03 (-0.16, 0.21)	0.29	1.42	0%
	Follow-up	3	387	0.04 (-0.16, 0.24)	0.4	0.21	0%
Quality of life	Post-treatment	3	136	0.05 (-0.29, 0.40)	0.29	0.98	0%
	Follow-up	3	123	-0.01 (-0.36, 0.38)	0.06	1.62	0%
Treatment satisfaction	Post-treatment	3	415	-0.39 (-0.71, -0.08)	2.42*	3.9	49%
	Follow-up	2	352	-0.35 (-0.56, -0.14)	3.22**	0.92	0%
Mixed Pain Conditions							
Pain Intensity	Post-treatment	4	410	-0.37 (-0.86, 0.11)	1.5	12.31	76%
	Follow-up	2	316	-0.45 (-1.68, 0.79)	0.71	13.84	93%
^a Disability	Post-treatment	2	317	-0.33 (-1.09, 0.42)	0.86	5.75	83%
^a Depression	Post-treatment	2	317	0.04 (-0.18, 0.26)	0.34	0.57	0%
^a Quality of life	Post-treatment	2	93	-0.07 (-0.48, 0.34)	0.34	0.66	0%

Data available only at post-treatment. † $p < 0.10.$, * $p < 0.05.$, ** $p < 0.01.$, *** $p < 0.001$

3.6.21 Adverse events

None of the included studies reported adverse events which may be attributed to study procedures. One study (Palermo et al., 2016) notes major life stressors and life events which account for the dropout of four participants. The authors also report an additional concern relating to risk of self-harm which was noted by the online coach associated with the trial. These events were reported as un-related to participation in the study. Explanation for participant dropouts were offered in detail by five studies (Cottrell et al., 2007; Connelly et al., 2006; Trautmann & Kröner-Herwig, 2010; Stinson et al., 2010a; Palermo et al., 2016), and in less detail by five other studies (McGrath et al., 1992; Hicks et al., 2006; Palermo et al., 2009; Rapoff et al., 2014; Law et al., 2015) which did not fully explain reasons for dropouts.

3.6.22 Sensitivity analysis

Of the trials included in meta-analyses, three trials showed a high risk of bias in three domains; blinding of participants and personnel, incomplete outcome data and selective reporting. We conducted a sensitivity analysis of the included trials where incomplete outcome data and selective reporting were rated as low risk of bias versus unclear or with a high risk of bias. We found that the effect of ICT-based therapies on headache reduction (OR = 4.03; 95% CI [1.50, 10.80], $I^2 = 36\%$) remained significant ($p < 0.01$) at post-treatment and had similar effect at follow-up (OR = 1.93; 95% CI [0.89, 4.23], $I^2 = 0\%$). Effects on pain intensity, disability and depression were unaltered. Due to the small number of trials we were unable to conduct a sensitivity analysis of the trials where blinding of participants and personnel rated as low risk of bias could be compared with trials which were scored as unclear or with a high risk of bias.

In addition to the analyses set out in the protocol, we conducted further sensitivity analyses of trials where intent to treat data analyses were not reported compared to trials which report intent to treat analyses. We found that

the effect of ICT-based therapies on headache reduction (OR = 5.09; 95% CI [1.63, 15.89], $I^2 = 51%$) remained significant ($p < 0.01$) at post-treatment and had similar effect at follow-up (OR = 1.93; 95% CI [0.89, 4.23], $I^2 = 0%$). Effects on pain intensity (SMD = -0.37; 95% CI [-0.86, 0.11]; $I^2 = 76%$); disability (SMD = -0.12; 95% CI [-0.44, 0.20]; $I^2 = 52%$); and depression (SMD = 0.01; 95% CI [-0.19, 0.20]; $I^2 = 0%$) remained non-significant ($p > 0.05$).

3.6.23 Subgroup analysis of intervention characteristics

As per our protocol, we planned subgroup analyses to investigate difference in effect between trials based on technology type (e.g. Internet versus telephone), contact with therapist (e.g. number/total contact with therapist) and pain type (e.g. headache pain, musculoskeletal or abdominal pain). However, due to the small number of trials we were unable to conduct these analyses. Post-hoc subgroup analyses were performed on included trials based on loss-to-follow-up. No significant differences were observed when endpoint numbers of drop-outs from the intervention and control conditions were compared (see Table 3.7).

Table 3.7

Subgroup analyses of loss to follow-up for headache and mixed pain conditions at each assessment point (treatment versus control).

Assessment point	<i>k</i>	<i>N</i>	Hedges <i>g</i> (95% CI)	<i>Z</i>	<i>Q</i>	<i>I</i> ²
Headache Pain Conditions						
Pre-enrolment	2	341	1.78 (0.78, 4.07)	1.37	0.2	0%
Post-treatment	7	591	1.39 (0.82, 2.37)	1.22	3.28	0%
Follow-up	5	437	0.68 (0.33, 1.39)	1.06	2.49	0%
Mixed Pain Conditions^a						
Pre-enrolment	2	319	2.83 (0.66, 12.19)	1.4	0.12	0%
Post-treatment	4	405	1.11 (0.30, 4.04)	0.15	4.56	34%

^a Data available only at post-treatment. † $p < 0.10$., * $p < 0.05$., ** $p < 0.01$., *** $p < 0.001$

Subgroup analyses were performed using studies that compared an intervention against a passive control group (i.e., either ‘WLC’ or minimal ‘attention control’) or active treatment group (see Figure 3.19). We found that the effect of ICT-based therapies on reduction in headache remained significant ($p < 0.001$) when compared with passive control conditions (OR = 8.29; 95% CI [3.01, 22.81], $I^2 = 0\%$) and active control conditions (OR = 2.23; 95% CI [1.18 4.25], $I^2 = 0\%$) at post-treatment.

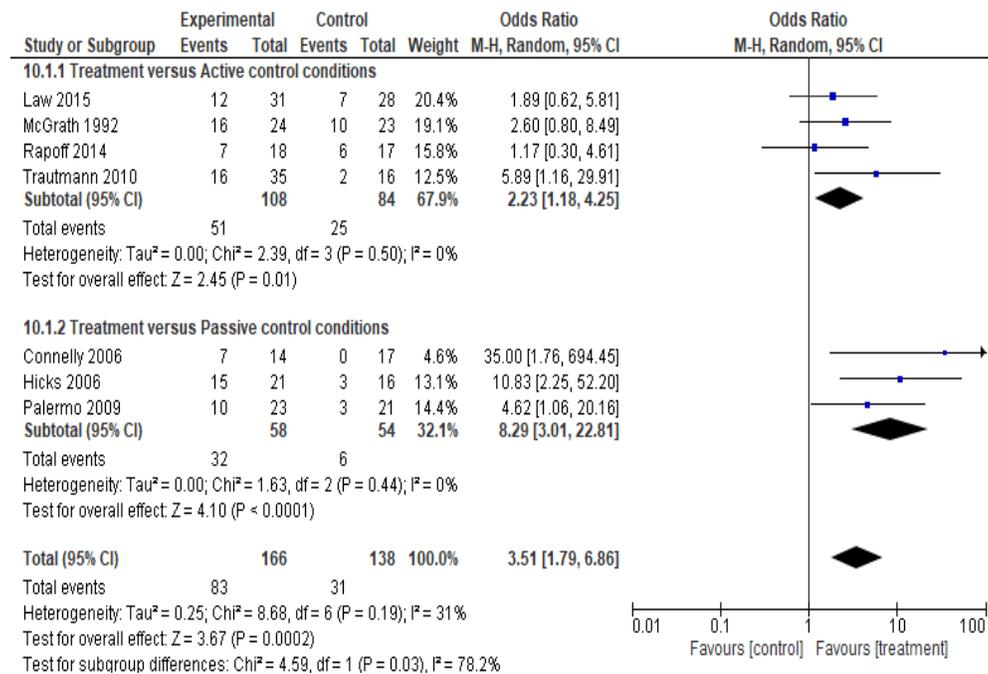


Figure 3.19 Subgroup analyses of CBT versus passive (WLC and passive ‘attention control’) comparison conditions for reduction in headache severity at post treatment.

At follow-up, we found the effect of ICT-based therapies on reduction in headache was non-significant ($p > 0.05$) when compared with only passive control conditions (OR = 4.41; 95% CI [0.42, 45.82], $I^2 = 76\%$) and only active control conditions (OR = 2.05; 95% CI [0.77, 5.43], $I^2 = 0\%$) (see Figure 3.20).

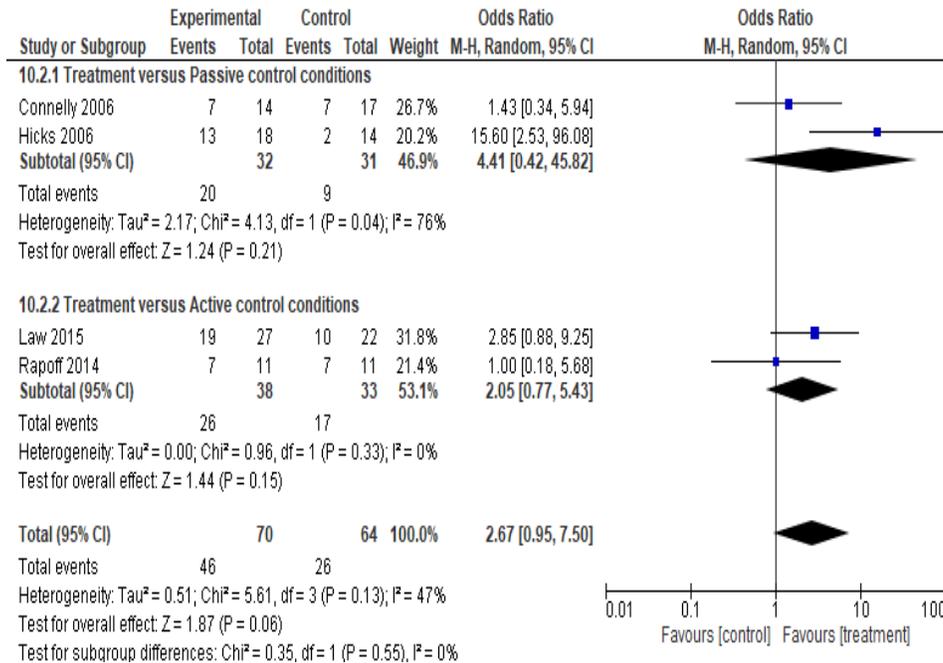


Figure 3.20 Subgroup analyses of CBT versus passive (WLC and passive ‘attention control’) comparison conditions for reduction in headache severity at follow-up.

We also found that the effect of ICT-based therapies on disability remained non-significant ($p > 0.05$) when compared with only the waitlist control conditions (OR = -0.33; 95% CI [-1.22, 0.55], $I^2 = 76\%$) and only the active control conditions (OR = -0.03; 95% CI [-0.23, 0.17], $I^2 = 0\%$) at post-treatment (see Figure 3.21). Analyses at follow-up could not be conducted because there were not enough interventions in the subgroup to allow for comparison. Due to the small number of included studies we were unable to run these analyses for other outcomes.

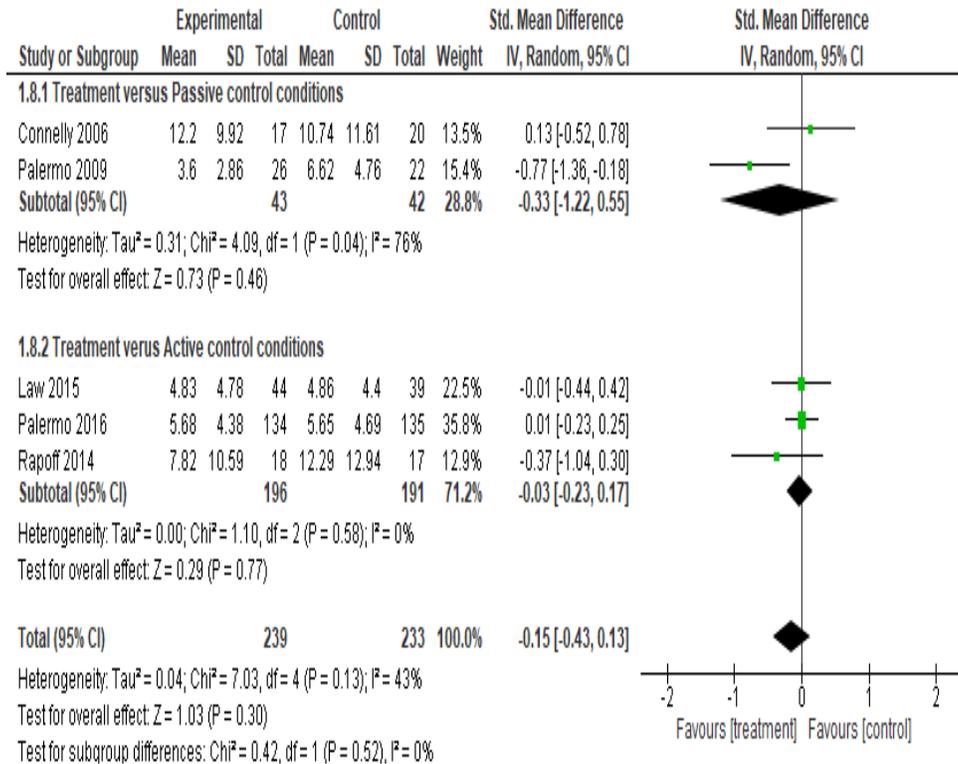


Figure 3.21 Subgroup analyses of CBT versus passive (WLC and passive ‘attention control’) comparison conditions for disability at post-treatment.

Finally, we planned to determine the difference in effect between trials that include or exclude various intervention characteristics including theoretical constructs, behaviour change techniques and usability factors. Tables 3.15, 3.21 and 3.22 present subgroup analyses of study characteristics, behaviour change techniques and theoretical domains across outcomes in each of the ten included trials.

3.6.24 The Theoretical Domains Framework

The presence of theoretical domains in the intervention arm(s) of included trials based on the Theoretical Domains Framework (Cane et al., 2012) is presented in Table 3.8.

Table 3.8

Presence of Theoretical Domains Framework domains in included studies

	Connolly 2006	Cottrell 2007	Hicks 2006	Law 2015	McGrath 1992	Palermo 2016	Palermo 2009	Rapoff 2014	Stinson 2010	Trautmann 2010
Knowledge	X	X	X	X	X	X	X	X	X	X
Skills	X	X	X	X	X	X	X	X	X	X
Social role and identity										
Beliefs about capabilities	X	X	X	X	X	X	X	X	X	X
Optimism										
Beliefs about consequences	X	X	X	X	X	X	X	X	X	X
Reinforcement						X	X			
Intentions										
Goals			X	X		X			X	
Memory, attention and decision processes	X	X	X	X	X	X	X	X	X	X
Environmental context and resources	X	X	X	X	X	X	X	X	X	X
Social influences	X	X	X	X	X	X	X	X	X	X

Emotion		X				X	X		X	X
Behaviour regulation	X	X	X	X	X	X	X	X	X	X

Of the fourteen domains within the Theoretical Domains Framework, the most frequently coded domain across ICT-based interventions was “knowledge” (10 out of 10 interventions, coded 37 times). In descending order, this is followed by “skills” (10 out of 10 interventions, coded 32 times); “behaviour regulation” (10 out of 10 interventions, coded 15 times), “memory, attention and decision processes” (10 out of 10 interventions, coded 14 times) and “social influences” (10 out of 10 interventions, coded 12 times). This is followed by “beliefs about consequences”, beliefs about capabilities” and “environmental contact and resources” which were each coded ten times in all ten interventions. The remaining domains were coded 5 or less times. These include, “goals” (4 out of 10 interventions, coded 5 times), “emotion” (5 out of 10 interventions, coded 5 times) and “reinforcement” (2 out of 10 interventions, coded twice) coded in 4 interventions. Finally, three domains were never coded: “social role and identity”, “intentions” and “optimism”. The number of times each theoretical domain was coded in the intervention group of each of the studies is shown in Table 3.9.

Table 3.10 presents the number of times each of the domains were coded in the control group of each of the trials. We identified and coded domains for nine of the control conditions. One control condition (Hicks et al 2006) was described simply as “standard medical care” with no further description. No domains were coded within this control condition. The most frequently coded domains across control conditions were “knowledge” (6 of 10 control conditions; coded 9 times) “social influences” (7 of 10 control groups; coded 7 times), “memory, attention and decision processes” (6 of 10 control conditions; coded 6 times) and “behaviour regulation” (4 of 10 control conditions, coded 4 times). “Skills”, and “environmental context and resources” were coded two or less times.

Table 3.9

The number of times each of the TDF domains were coded in the intervention group of the included studies.

Domains	Connolly 2006 CBT	Cottrell 2007 CBT	Hicks 2006 CBT	Law 2015 CBT	McGrath 1992 CBT	Palermo 2016 CBT	Palermo 2009 CBT	Rapoff 2014 CBT	Stinson 2010 CBT	Trautmann 2010 CBT&AR	Total
1. Knowledge	4	5	5	3	1	4	3	4	6	2	37
2. Skills	3	4	3	5	5	2	4	1	3	2	32
3. Social role and identity	0	0	0	0	0	0	0	0	0	0	0
4. Beliefs about capabilities	1	1	1	1	1	1	1	1	1	1	10
5. Optimism	0	0	0	0	0	0	0	0	0	0	0
6. Beliefs about consequences	1	1	1	1	1	1	1	1	1	1	10
7. Reinforcement	0	0	0	0	0	1	1	0	0	0	2
8. Intentions	0	0	0	0	0	0	0	0	0	0	0
9. Goals	0	0	2	1	0	1	0	0	1	0	5
10. Memory, attention and decision processes	2	1	1	2	1	1	2	1	2	1	14
11. Environmental context and resources	1	1	1	1	1	1	1	1	1	1	10
12. Social influences	1	1	1	1	1	3	1	1	1	1	12

13. Emotion	0	1	0	0	0	1	1	0	1	1	5
14. Behaviour regulation	2	2	2	0	2	3	1	1	2	0	15
Total number of domains targeted	15	17	17	15	13	19	16	11	19	10	

CBT: cognitive behavioural therapy; AR: applied relaxation

Table 3.10

Number of times each of the TDF domains were coded in the control group of each of the included studies.

Domains	Connolly 2006	Cottrell 2007	Hicks 2006	Law 2015	McGrath 1992	Palermo 2016	Palermo 2009	Rapoff 2014	Stinson 2010	Trautmann 2010
1. Knowledge	1	0	0	1	1	1	0	3	0	2
2. Skills	0	0	0	1	1	0	0	0	0	0
3. Social role and identity	0	0	0	0	0	0	0	0	0	0
4. Beliefs about capabilities	0	0	0	0	0	0	0	0	0	0
5. Optimism	0	0	0	0	0	0	0	0	0	0
6. Beliefs about consequences	0	0	0	0	0	0	0	0	0	0
7. Reinforcement	0	0	0	0	0	0	0	0	0	0
8. Intentions	0	0	0	0	0	0	0	0	0	0
9. Goals	0	0	0	0	0	0	0	0	0	0
10. Memory, attention and decision processes	1	1	0	1	0	1	1	0	1	0
11. Environmental context and resources	0	0	0	0	0	0	0	1	0	0
12. Social influences	1	1	0	1	1	0	0	1	1	1
13. Emotion	0	0	0	0	0	0	0	0	0	0

14. Behaviour regulation	0	0	0	0	1	1	1	0	0	1
Total number of domains targeted	3	2	0	4	4	3	2	5	2	4

CBT: cognitive behavioural therapy; AR: Applied relaxation

Relationship between total number of times the domains are coded within an intervention and effect size.

Table 3.11 summarises the total number of times any of the domains were coded, for each study intervention and control group.

Table 3.11
*Total number of times **any** domain was coded within intervention and control groups*

Studies	Total no any domain coded Intervention	Total no any domain coded Control	Intervention minus Control
Connolly 2006	15	3	12
Cottrell 2007	18	1	17
Hicks 2006	17	0	17
Law 2015	16	4	12
McGrath 1992	14	4	10
Palermo 2016	20	3	17
Palermo 2009	17	2	15
Rapoff 2014	11	6	5
Stinson 2010	18	2	16
Trautmann 2010	17	5	12

The relationship between the total number of times the domains were coded within an intervention and post-treatment estimates of effect were non-significant ($p > 0.05$). Sensitivity analysis (before subtraction of control group domains) did not alter this result ($p > 0.05$) (see Table 3.12).

Table 3.12
Relationship between the total number of times the domains were coded within an intervention and effect size

Outcome	<i>n</i>	<i>r</i>	<i>p</i>	Pre-sensitivity analyses*	
				<i>r</i>	<i>p</i>
Reduction in headache pain	328	-0.23	0.62	0.12	0.79
Disability	472	-0.15	0.81	-0.32	0.60
Depression	455	0.37	0.63	-0.10	0.90
Quality of life	182	0.42	0.58	0.41	0.59
Pain intensity	410	0.86	0.14	0.51	0.49

*relationship before subtracting the number of control group domains

Relationship between the number of different domains coded within an intervention and the effect size.

Table 3.13 summarises the total number of times any of the domains were coded, for each study intervention and control group.

Table 3.13
*Total number of times **different** domains were coded within intervention and control groups*

Studies	Total no of different domains coded in Intervention	Total no of different domains coded Control	Intervention minus Control
Connolly 2006	8	3	5
Cottrell 2007	9	2	7
Hicks 2006	9	0	9
Law 2015	8	4	4
McGrath 1992	8	4	4
Palermo 2016	11	3	8
Palermo 2009	10	2	8
Rapoff 2014	8	3	5
Stinson 2010	10	2	8
Trautmann 2010	8	3	5

The relationship between the total number of times different domains which were coded within an intervention and post-treatment estimates of effect

were not significant ($p > 0.05$). Sensitivity analysis (no subtraction of control group domains) showed similar results ($p > 0.05$) (see Table 3.14).

Table 3.14

Relationship between the total number of different domains coded within an intervention and effect size

Outcome	<i>n</i>	<i>r</i>	<i>p</i>	Pre-sensitivity analyses*	
				<i>r</i>	<i>p</i>
Reduction in headache pain	328	-0.06	0.9	-0.11	0.81
Disability	472	-0.46	0.44	-0.61	0.27
Depression	455	-0.14	0.86	-0.14	0.86
Quality of life	182	-0.32	0.69	0.15	0.85
Pain intensity	410	0.74	0.26	-0.23	0.77

*relationship before subtracting the number of control group domains

Only two trials mentioned the use of theory as a guiding framework (Palermo et al., 2009; Palermo et al., 2016) however neither trial described how the theory was implemented in intervention development or selection of participants. Although the interventions were informed by Social Cognitive Theory (Bandura, 1986; 1991), Social Learning Theory (Bandura, 1977) and Family Systems Theory (Kerr, 2000) the expected theoretical constructs were not always targeted or measured. Studies that made explicit reference to a theoretical framework were not associated with substantially larger effects relative to interventions which failed to report or use an underlying theoretical framework. Also, interventions that targeted a greater number of theoretical domains did not tend to have larger effects on behaviour than did interventions that targeted fewer theoretical domains. We found no association between the total number of theoretical domains identified and estimates of effect on headache severity ($r = .122$, $p = .79$), pain intensity ($r = .856$, $p = .14$) or disability ($r = .163$, $p = .79$), depression ($r = -0.99$, $p = .08$), quality of life ($r = .42$, $p = .58$) or treatment satisfaction ($r = -.80$, $p = .41$), across headache and mixed pain outcomes at post-treatment.

Subgroup meta-analysis of pain, functioning and psychosocial outcomes in response to ICT-based psychology therapies were carried out based on the presence or absence of specific TDF domains which were targeted by intervention and control groups and reported in at least two RCTs. The small number of studies meant that for most domains there were not enough interventions to allow for subgroup comparison. Where sufficient data were available we compared estimates of effect in response to ICT-based psychology therapies based on the presence or absence of three theoretical domains; ‘Goals’ ‘Reinforcement’ and ‘Emotion’ (see Table 3.15 and Figure 3.22).

- a) *Reduction in headache severity.* Analyses of headache reduction in response to treatment based on the presence or absence of TDF categories revealed no significant differences in estimates of effect between those studies that targeted or did not target ‘Reinforcement’ ($t(2) = 1.000, p = 0.42$) or ‘Goals’ ($t(1.2) = 0.422, p = 0.74$). Insufficient evidence prevented comparisons based on ‘Emotion’.
- b) *Pain Intensity.* Analyses of pain intensity in response to treatment based on the presence or absence of TDF categories revealed no significant differences in estimates of effect between those studies targeting or not targeting ‘Reinforcement’ ($t(1.51) = 1.457, p = 0.32$), ‘Goals’ ($t(2) = .126, p = 0.91$) or ‘Emotion’ ($t(2) = .213, p = 0.85$).
- c) *Disability.* Analyses of disability in response to treatment based on the presence or absence of these TDF categories revealed no significant differences between those studies targeting or not targeting ‘Reinforcement’ ($t(1.2) = -1.012, p = 0.48$), ‘Goals’ ($t(1.02) = -.612, p = 0.65$) and ‘Emotion’ ($t(1.16) = -1.012, p = 0.48$).
- d) *Depression.* Analyses of depression in response to treatment revealed no significant differences between in estimates of effect between those studies targeting or not targeting ‘Reinforcement’ ($t(1.85) = -.211, p = 0.85$), ‘Goals’ ($t(1.84) = -.200, p = 0.86$) or ‘Emotion’ ($t(2) = -.616, p = 0.60$).
- e) *Quality of life.* Analyses of quality of life in response to treatment revealed

no significant differences in estimates of effect between those studies targeting or not targeting ‘Goals’ ($t(1.79) = -.292, p = 0.80$) and ‘Emotion’ ($t(1.01) = -5.161, p = 0.12$) and insufficient evidence to allow comparison based on ‘Reinforcement’.

- f) *Treatment satisfaction*. There was insufficient data to allow subgroup comparison.

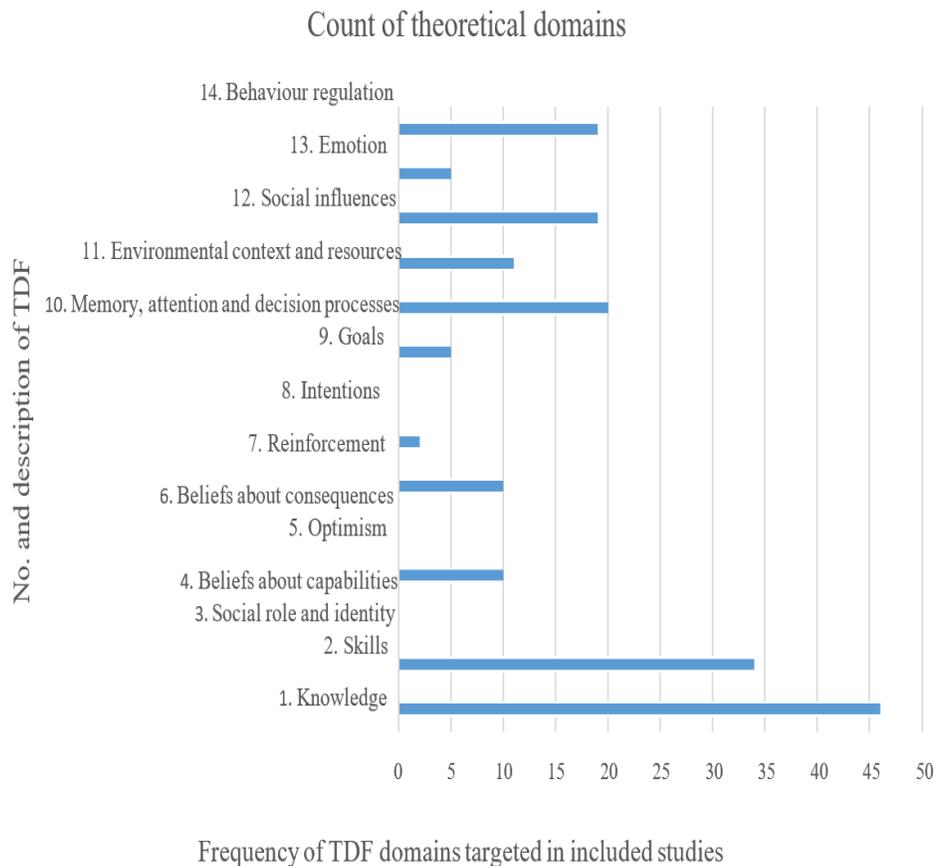


Figure 3.22 Count of TDF Framework

Table 3.15

Meta-analyses and t-test results for comparison of intervention effects on outcomes for studies including/excluding TDFs.

TDF Domains	TDF domains applied				TDF domains not applied				t	df	p
	k	N	OR ^a /SMD	95% CI	k	N	ORa/SMD	95% CI			
Reduction in headache^a											
TDF7 Reinforcement	2	91	3.26	1.29, 8.19	5	237	3.86*	1.36, 10.92	1	2	0.42
TDF9 Goals	2	120	3.91	0.65, 23.62	5	208	3.39**	1.49, 7.69	0.422	1.2	0.74
TDF13 Emotion	2	95	5.15	1.73, 15.36	5	233	3.13*	1.24, 7.88	n/a		
Disability											
TDF7 Reinforcement	2	317	-0.33	-1.09, 0.42	3	155	-0.05	-0.37, 0.26	-0.01	1.16	0.48
TDF9 Goals	2	352	0	-0.21, 0.21	3	120	-0.35	-0.87, 0.17	-0.61	1.02	0.65
TDF13 Emotion	2	317	-0.33	-1.09, 0.42	3	155	-0.05	-0.37, 0.26	-0.01	1.16	0.48
Depression											
TDF7 Reinforcement	n/a	0.211	1.85	0.85							

TDF9 Goals	n/a	-0.2	1.84	0.86
TDF13 Emotion	n/a	-0.62	2	0.6

OR^a: Odds ratio. n/a = not enough interventions in the subgroup to allow for comparison.

Table 3.15

Meta-analyses and t-test results for comparison of intervention effects on outcomes for studies including/excluding TDFs continued....

TDF Domains	TDF domains applied				TDF domains not applied				t	df	p
	k	N	ORa/SMD	95% CI	k	N	ORa/SMD	95% CI			
Quality of life											
TDF7 Reinforcement	n/a										
TDF9 Goals	2	93	0	-0.46, 0.46	2	89	0.03	-0.41, 0.46	-0.29	1.79	0.8
TDF13 Emotion	2	100	-0.01	-0.46, 0.43	2	82	0.04	-0.40, 0.47	-5.16	1.01	0.12
Pain intensity											
TDF7 Reinforcement	2	317	-0.16	-0.82, 0.51	2	93	-0.63**	-1.05, -0.21	1.457	1.52	0.32
TDF9 Goals	n/a										
TDF13 Emotion	n/a										

OR^a: Odds ratio. n/a = not enough interventions in the subgroup to allow for comparison.

3.6.25 Behaviour Change Techniques

Figure 3.23 displays the frequency of inclusion of the different BCTs. Table 3.16 shows the presence of behaviour change techniques in the intervention arm(s) of included trials based on v1 of the Behaviour Change Taxonomy (Michie et al., 2013). Twenty-one of the 93 BCTs in version 1 of the Behaviour Change taxonomy were identified in the studies reviewed.

The most commonly reported BCTs in the intervention conditions were: 4.1 instruction on how to perform behaviour and 12.6 body changes ($k = 10$). The second most frequently coded BCT's were 1.2 problem solving and 8.1 behavioural practice/rehearsal ($k = 8$). This is followed by 3.1 social support (unspecified), 6.1 demonstration of the behaviour, 11.2 reduce negative emotions, 13.2 framing/re- framing which were coded in five interventions. The most commonly reported BCTs in the control conditions were: 11.1 pharmacological support ($k = 3$), 4.2 information about antecedents ($k = 3$), 3.1 social support (unspecified), the remaining BCTs were coded once ($k = 2$), 2.6 biofeedback, 5.3 information about social and environmental consequences, 7.1 prompts/cues and 9.1 credible source were coded only once ($k = 2$). The number of incorporated BCTs per trial ranged from 8 to 19 with a median of 11 (11.3 mean). The present review found no evidence to suggest interventions that made extensive use of BCTs are associated with greater reductions in pain severity ($r = -.31, p = .50$), disability ($r = .26, p = .67$), depression ($r = -.18, p = .82$), quality of life ($r = .27, p = .83$), treatment satisfaction ($r = .40, p = .74$) and pain intensity ($r = .35, p = .77$) across headache and mixed pain outcomes or at follow-up.

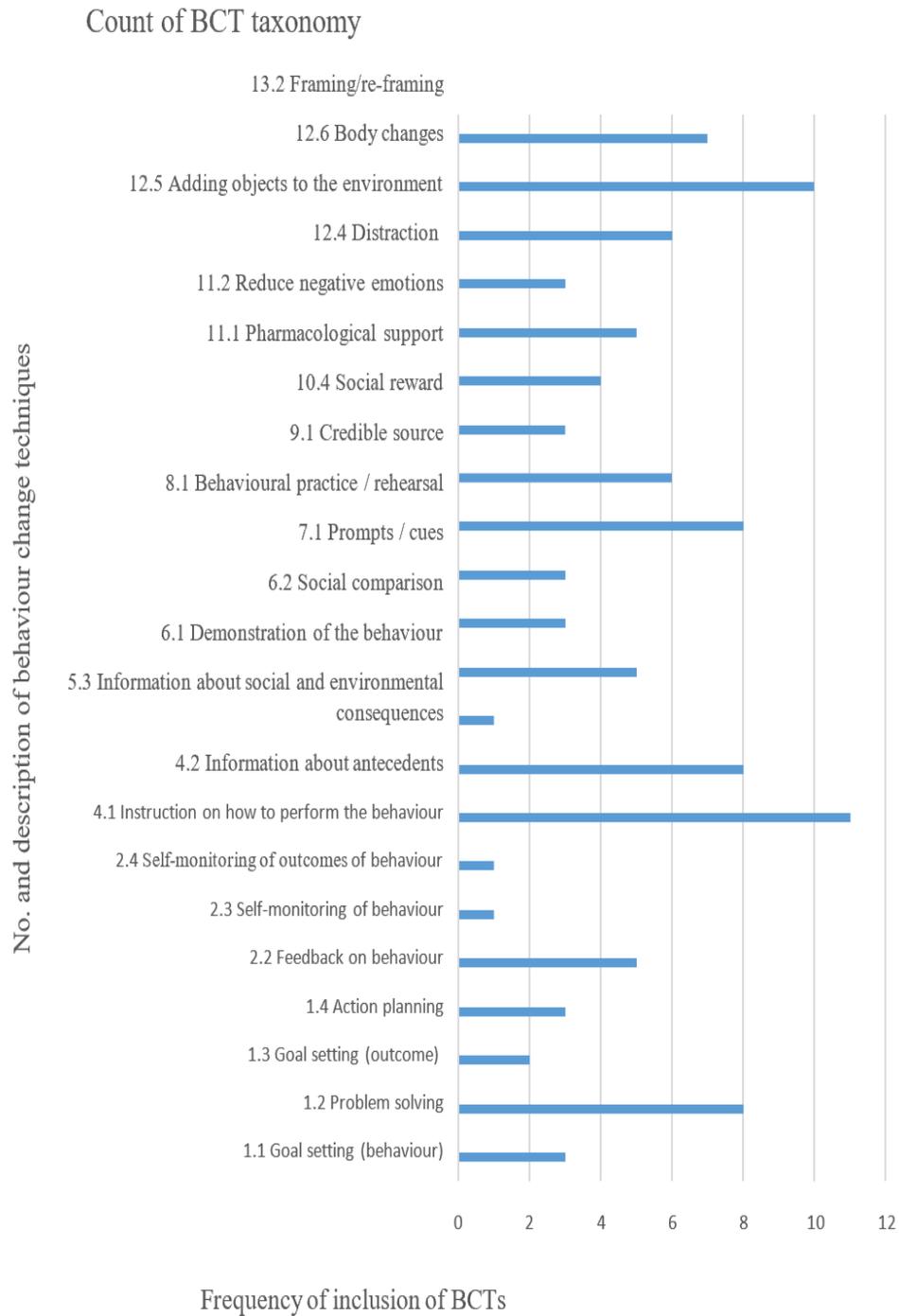


Figure 3.23 Count of BCT taxonomy.

Table 3.16

Presence of behaviour change techniques (v1 The Behaviour Change Taxonomy)

	Connolly 2006	Cottrell 2007	Hicks 2006	Law 2015	McGrath 1992	Palermo 2009	Palermo 2016	Rapoff 2014	Stinson 2010	Trautmann 2010
1.1 Goal setting (behaviour)			X	X			X			
1.2 Problem solving	X	X	X	X	X	X		X		X
1.3 Goal setting (outcome)						X			X	
1.4 Action planning	X		X				X			
2.2 Feedback on behaviour		X	X	X		X	X			
2.3 Self-monitoring of behaviour							X			
2.4 Self-monitoring of outcomes of behaviour	X									
2.6 Biofeedback		X		X						
3.1 Social support (unspecified)		X	X		X	X			X	
3.2. Social support (practical)		X								
3.3 Social support (emotional)							X			
4.1 Instruction on how to perform the behaviour	X	X	X	X	X	X	X	X	X	X
4.2 Information about antecedents			X							X

5.3 Information about social and environmental consequences			X							
6.1 Demonstration of the behaviour	X			X		X	X	X		
6.2 Social comparison			X	X			X			
7.1 Prompts / cues										
8.1 Behavioural practice / rehearsal		X	X	X	X	X	X	X	X	X
9.1 Credible source		X				X	X			
10.4 Social reward			X			X	X			
11.1 Pharmacological support		X								
11.2 Reduce negative emotions	X	X				X	X	X		X
12.4 Distraction				X	X				X	
12.5 Adding objects to the environment		X	X		X			X	X	X
12.6 Body changes	X	X	X	X	X	X		X	X	
13.2 Framing/re-framing	X		X	X	X	X	X	X		X
15.4 Self-talk			X							

Table 3.17 summarises the total number of times any of the BCTs were coded, for each study intervention and control group.

Table 3.17

Total number of times any BCTs were coded in the intervention and control group of each study

Studies	Total no any BCT coded	Total no any BCT coded Control	Intervention minus Control
	Intervention		
Connolly 2006	8	1	7
Cottrell 2007	12	1	11
Hicks 2006	15	0	15
Law 2015	12	3	9
McGrath 1992	8	2	6
Palermo 2009	11	1	10
Palermo 2016	13	2	11
Rapoff 2014	8	2	6
Stinson 2010	8	1	7
Trautmann 2010	8	1	7
Total	103	14	89

Table 3.18 summarises the total number of times different BCTs were coded, for each study intervention and control group.

Table 3.18

*Number of times **different** BCTs were coded in the intervention and control group of each study*

Behaviour change techniques	Total no different BCT coded in intervention groups	Total no different BCT coded in control groups	Intervention minus control
1.1	3	0	3
1.2	8	0	8
1.3	2	0	2
1.4	3	0	3
2.2	5	0	5
2.3	1	0	1
2.4	1	0	1
4.1	11	0	11
4.2	2	6	-4
5.3	1	0	1
6.1	5	0	5
6.2	3	0	3
7.1	2	1	1
8.1	8	0	8
9.1	3	3	0
10.4	3	0	3
11.1	2	2	0
11.2	5	0	5
12.4	3	0	3
12.5	5	1	4
12.6	10	0	10
13.2	7	0	7
Total	86	13	73

Sensitivity analysis (no subtraction of control group BCTs) did not alter these findings at post-treatment or follow-up ($p > 0.05$) (see Table 3.19 and 3.20).

Table 3.19

Relationship between the total number of BCTs coded within an intervention and effect size (post-treatment)

Pre-sensitivity analysis*					
Outcome	<i>n</i>	<i>r</i>	<i>p</i>	<i>r</i>	<i>p</i>
Headache pain conditions					
Reduction in headache pain	328	-0.31	0.5	-0.3	0.52
Disability	472	0.26	0.67	0.01	0.99
Depression	455	-0.18	0.82	-0.33	0.68
Quality of life	136	0.27	0.83	0.16	0.9
Treatment satisfaction	415	0.4	0.74	0.35	0.77
Mixed pain conditions					
Pain intensity	410	0.88	0.12	0.66	0.34
Disability	n/a				
Depression	n/a				
Quality of life	n/a				

*no subtraction of control group BCTs, n/a = not enough interventions to allow correlation

Subgroup meta-analysis of pain, functioning and psychosocial outcomes in response to ICT-based psychology therapies were carried out based on the presence or absence of specific BCTs which were reported in at least two RCTs (Table 3.20).

- a) *Pain intensity*. Subgroup analyses revealed no significant differences between studies including or excluding the use of the BCTs ‘problem solving’, ‘goal setting (behaviour)’, ‘reduce negative emotions’, ‘social comparison’ ‘action planning’ and ‘demonstration of behaviour’.
- b) *Reduction in headache severity*. Subgroup analyses revealed no significant differences between those studies including or excluding the use of the BCTs ‘distraction’, ‘adding objects to the environment’, ‘action planning’, ‘feedback on behaviour’, goal setting (behaviour), ‘social reward’, ‘social comparison’ ‘reduce negative emotions’ and credible source’. However, there was a significant difference between studies which incorporate BCT 6.1 ‘demonstration of the behaviour’ ($t(3) = 5.3, p = 0.01$).
- c) *Disability*. Subgroup analyses of disability revealed no significant differences between those studies including or excluding the use of the BCTs ‘action planning’, ‘feedback on behaviour’, ‘goal setting

(behaviour)', 'social reward', 'social comparison' 'reduce negative emotions' and credible source'.

- d) *Depression*. Subgroup analyses revealed no significant differences between those studies including or excluding use of the BCTs 'social reward', 'social comparison' and credible source'.
- e) *Quality of life*. Subgroup analyses revealed no significant differences between those studies including or excluding use of the BCT 'information about antecedents'.
- f) *Treatment satisfaction*. There was insufficient data to allow subgroup comparison.

Table 3.20

Subgroup analyses of intervention effects on outcomes for studies including/excluding BCTs

Reduction in headache	BCTs present				BCTs absent				<i>t</i>	<i>df</i>	<i>p</i>
	BCTs	k	N	OR/SMD	95% CI	k	N	OR/SMD			
BCT1.1	2	120	3.91	0.65, 23.62	2	208	3.39**	1.49, 7.69	0.7	5	0.54
BCT1.4	2	68	13.97***	3.47, 56.17	5	260	2.41**	1.35, 4.30	0.3	5	0.75
BCT 2.2	3	164	3.88*	1.31, 11.49	4	164	3.30*	1.13, 9.62	0.3	5	0.79
BCT4.2	2	88	8.07***	2.61, 24.97	5	240	2.50*	1.20, 5.21	1.8	5	0.13
BCT6.1	4	193	2.68*	0.96, 7.45	3	135	4.75***	2.04, 11.07	5.3	3	0.01
BCT6.2	2	120	3.91	0.65, 23.62	5	208	3.39**	1.49, 7.69	0.7	5	0.54
BCT10.4	2	81	6.88***	2.35, 20.17	5	247	2.61*	1.19, 5.72	0.3	5	0.81
BCT11.2	3	130	2.98*	1.08, 8.23	4	198	4.18**	1.42, 12.34	0.5	5	0.67
BCT12.4	2	130	2.06 †	0.94, 4.53	5	198	4.96**	1.86, 13.27	0.9	5	0.42
BCT12.5	4	170	3.44**	1.37, 8.66	3	158	3.97*	1.02, 15.42	1.4	5	0.24
Disability	BCTs present				BCTs absent						
BCT1.1	2	352	0	-0.21, 0.21	3	120	-0.35	-0.87, 0.17	1.2	3	0.31
BCT1.4	2	306	0.02	-0.20, 0.25	3	166	-0.35	-0.82, 0.12	0.8	3	0.48
BCT 2.2	3	400	-0.19	-0.59, 0.21	2	72	-0.11	-0.60, 0.38	2.3	1	0.23
BCT6.2	2	352	0	-0.21, 0.21	3	120	-0.35	-0.87, 0.17	0.8	3	0.48
BCT9.1	2	317	-0.33	-1.09, 0.42	3	155	-0.05	-0.37, 0.26	1.3	3	0.29
BCT10.4	2	317	-0.33	-1.09, 0.42	3	155	-0.05	-0.37, 0.26	1.3	3	0.29

BCT11.2	3	352	-0.32	-0.82, 0.18	2	120	0.04	-0.32, 0.39	0.9	3	0.44
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OR^a: Odds ratio effect estimate, Analyses of some BCT's could not be evaluated due to insufficient number of trials included in measures, † p < 0.10, n/a = not enough interventions in the subgroup to allow for a comparison.

Table 3.20

Subgroup analyses of intervention effects on outcomes for studies including/excluding BCTs

Depression	BCTs present				BCTs absent						
BCT6.2	2	352	0.03	-0.18, 0.24	2	103	0.02	-0.38, 0.42	0.2	2	0.86
BCT9.1	2	317	0.04	-0.18, 0.26	2	138	0	-0.34, 0.34	0.2	2	0.85
BCT10.4	2	317	0.04	-0.18, 0.26	2	138	0	-0.34, 0.34	0.2	2	0.85
Quality of life	BCTs present				BCTs absent						
BCT 2.3	2	101	0.22	-0.19, 0.63	2	81	-0.23	-0.67, 0.20	0.2	2	0.83
Pain intensity	BCTs present				BCTs absent						
BCT1.1	2	316	-0.16	-0.82, 0.51	2	94	-0.63	-1.05, -0.21	1.3	2	3.60
BCT1.2	2	95	-0.55**	-0.96, -0.14	2	315	-0.24	-1.07, 0.58	0.3	1	0.82
BCT1.4	2	316	-0.16	-0.82, 0.51	2	94	-0.63**	-1.05, -0.21	1.3	2	3.60
BCT6.1	2	316	-0.16	-0.82, 0.51	2	94	-0.63**	-1.05, -0.21	1.5	2	0.32
BCT6.2	2	316	-0.16	-0.82, 0.51	2	94	-0.63**	-1.05, -0.21	1.3	2	3.60
BCT11.2	2	316	-0.16	-0.82, 0.51	2	94	-0.63**	-1.05, -0.21	1.5	2	0.32

OR^a: Odds ratio effect estimate, Analyses of some BCT's could not be evaluated due to insufficient number of trials included in measures, † p < 0.10, n/a = not enough interventions in the subgroup to allow for a comparison.

3.6.26 Mode of delivery

The included ICT-based interventions used web-based ($k = 6$), CD-ROM ($k = 2$) and telephone treatment delivery formats ($k = 2$). All treatment interventions used the following modes of delivery: (a) providing an enriched information environment, (f) access to advisor to request advice, (g) scheduled contact with advisor and tunnelled navigational formats ($k = 10$). A total of nine treatment interventions provided (e) supported progress monitoring ($k = 9$), Half the included interventions used (i) email in addition to the Internet-based treatment delivery ($k = 5$), Only one intervention used (h) peer-to-peer access ($k = 1$). None of the included interventions reported the (b) provision of automated tailored feedback, (c) generic follow-up messages, (k) text-messaging, (m) video-conferencing, (q) free-choice navigational formats or referred to (t) explicit citing of credentials ($k = 10$), for mode of delivery coding sheet) (see *Figure 3.24*).

Like Webb et al (2010) we planned to analyse modes of delivery according to the function categories: automated functions, communicative functions, use of supplementary modes, navigational format, entertainment value and credibility in comparison with control condition. However, insufficient data prevented analyses across outcomes. Six out of nineteen modes of delivery are included in analyses and presented Table 3.21.

- a) *Entertainment value*: (Appearance; Mode S or quizzes, stories or graphics; Mode R) Interventions that emphasised ‘entertainment value’ modes of delivery were associated with significantly smaller estimates of effect for reduction in headache severity compared to interventions that did not contain these features ($t(3.4) = -5.25$, $p = 0.01$).
- b) *Supplementary modes of delivery*. Analyses revealed no significant differences between interventions that used supplementary email (Mode N) or supplementary ($t(5) = 1.09$, $p = 0.33$).
- c) *Mode E: Supported progress monitoring*. Analyses revealed no significant differences between interventions that offered supported progress monitoring and those that did not offer this mode of delivery ($t(4.9) = 0.47$, $p = 0.66$).

d) *Overall method of delivery.* Analyses revealed no significant differences between interventions that used computer (CD-ROM) or telephone-based platform ($t(5) = 1.09, p = .33$).

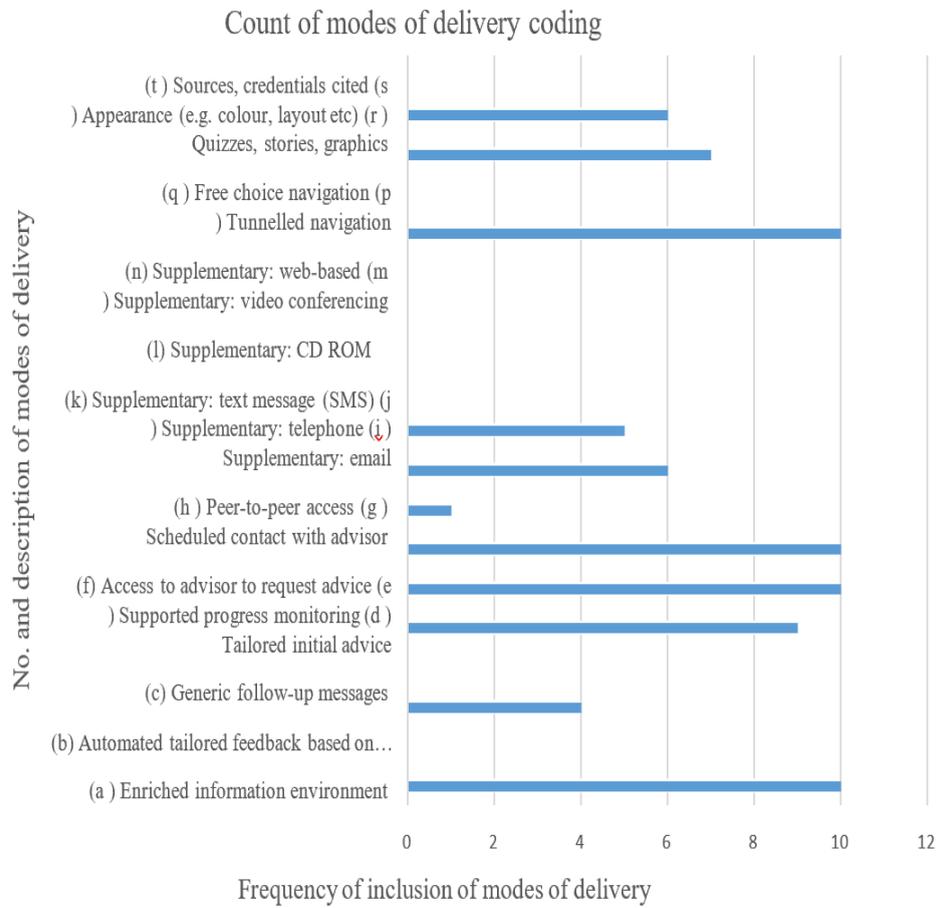


Figure 3.24 Count of modes of delivery.

Table 3.21

Meta-analyses and t-test results for comparison of intervention effects on outcomes for studies including/excluding modes of delivery.

Modes of delivery	Mode applied				Mode not applied				t	df	p
	k	N	ORa/SMD	95% CI	k	N	ORa/SMD	95% CI			
Reduction in headache severity											
Mode E	3	178	2.94	1.35, 6.40	4	152	4.3	1.28, 14.48	0.47	4.9	0.66
Mode J	3	115	3.1	0.75, 12.72	4	215	4.06	1.77, 9.34	1.09	5	0.33
Mode N	4	215	4.06	1.77, 9.34	3	115	3.1	0.75, 12.72	1.09	5	0.33
Mode R	4	193	2.68	0.96, 7.45	3	137	7.24	3.13, 16.73	-5.25	3.4	0.01
Mode S	4	193	2.68	0.96, 7.45	3	137	7.24	3.13, 16.73	-5.25	3.4	0.01
Disability											
Mode C	3	400	-0.19	-0.59, 0.21	2	72	-0.11	-0.60, 0.38	-2.34	1.2	0.23

OR^a: Odds ratio. Mode C: generic follow-up messages; Mode E: supported progress monitoring; Mode J: supplementary telephone, Mode N: supplementary email; Mode R: quizzes, stories graphics, Mode S: appearance

3.7 Discussion

3.7.1 Summary of findings

This is the first systematic review of the effectiveness of ICT-based psychological therapies for paediatric chronic pain which systematically coded behavioural intervention content based on theory, active ingredients and functionality. The present review involves separate meta-analyses, narrative synthesis and exploratory analyses of randomised controlled trials involving children with headache and mixed pain conditions. Narrative synthesis included ten trials (N=747) that delivered psychological therapy to children and adolescents using information and communication technology. Nine of these trials (N= 717) were included in quantitative synthesis. In line with previous reviews headache and mixed pain conditions (e.g. musculoskeletal pain, juvenile idiopathic arthritis and abdominal pain) were analysed separately (Eccleston et al., 2014b; Fisher et al., 2015). We examined the impact of ICT-based therapies on pain and disability in children and adolescents with chronic pain. We also attempted to assess the impact of ICT-based interventions on secondary outcomes including anxiety, depression, quality of life, parental protectiveness and treatment satisfaction. Exploratory analyses were focused on the relationship between estimates of effect, methodological and intervention content. Methodological quality was determined using Cochrane Collaboration recommended measures.

We found some evidence to suggest that ICT-based psychological therapies have a beneficial effect on reduction of headache severity after treatment. Results from the primary meta- analysis of end-point data showed a significant between group difference in reduction in headache severity (defined as >50% or more) at post-treatment but not at follow-up. Our results indicate the odds of achieving meaningful reduction in headache severity were 3.44 times higher for participants in the ICT-based intervention groups. These results are in accordance with previous reviews evaluating technology-based treatment (Fisher et al., 2015) and face to face psychological therapy (Eccleston et al., 2014b) for paediatric chronic pain. The post- treatment result (RR= 2.04; OR = 3.44) obtained was marginally

lower than the results found by Fisher et al (2015) (headache reduction: RR = 2.65) and Eccleston et al (2014) (RR = 2.47). According to the current findings approximately three children would need to receive treatment to receive benefit at post treatment (NNTB = 3.91), this figure is less optimistic compared to the NNTB reported by Fisher et al (2015) (NNTB = 2.88) and Eccleston et al 2014 (NNTB = 2.94).

No beneficial effects of ICT-based psychological therapy were found at post-treatment or follow-up assessment of pain intensity for children with mixed pain conditions. This contrasts with results reported by Fisher and colleagues (2015). The authors report significant reduction in pain intensity among children with mixed pain conditions at post-treatment. Differences in estimates of effect may be explained by the inclusion of an additional much larger, high quality trial (Palermo et al., 2016) in the present analysis. If this trial were removed, the effect of ICT-based treatment on pain intensity would be significant in favour of treatment.

No beneficial effects of ICT-based psychological therapy were found at post-treatment or follow-up assessment of disability. These results are consistent with the previous findings (Fisher et al., 2015). Fisher and colleagues report no beneficial effects of treatment on disability at post-treatment and insufficient data prevented follow-up analyses among headache and mixed pain conditions. The present review was able to conduct analyses at follow-up for children with headache pain but not mixed pain conditions.

No beneficial effects of ICT-based psychological therapy were found at post-treatment or follow-up assessment of depression or quality of life. This is in line previous findings (Fisher et al., 2015). Fisher and colleagues report no beneficial effects of treatment on depression across headache or mixed pain conditions. Follow-up analyses were prevented by insufficient data. The present review was able to conduct analyses of depression at follow-up for children with headache pain but not mixed pain conditions.

Treatment satisfaction in response to technology-based therapies has never been quantitatively evaluated for children with chronic or recurrent pain. Our results indicate that ICT-based psychological therapies are associated with significant treatment satisfaction among children with chronic or recurrent headache pain. Narrative syntheses have previously been prevented by the heterogeneity of instruments used to measure this outcome and by individual trials which fail to measure treatment satisfaction across trials arms (Fisher et al., 2015).

3.7.2 Potential moderators of treatment effect

In the current review, treatment effects varied widely between studies. Subsequent subgroup meta-analyses suggest trial design characteristics may significantly moderate the beneficial effects of ICT-based therapies on headache reduction.

Interventions which were compared to only waitlist control conditions (OR = 6.12; RR = 3.56) or waitlist and passive, attention control conditions (OR = 7.25; RR = 4.02) had higher odds of achieving significant reduction in headache severity than the interventions which were compared to active, education-based control conditions (OR = 2.78; RR = 1.90). While this was not maintained at follow-up, this finding could suggest an over-estimation of treatment effects which is specific to ICT-based psychological therapy versus waiting-list control conditions. These comparisons are limited by the small number of trials included in analyses and by moderate heterogeneity observed in the active comparison subgroup. Other potential moderators which might explain part of this result include the level of training and experience of the providers who offer supplemental social support in conjunction to treatment or the functionality of the overall intervention.

Sensitivity analyses were performed as planned. No additional or significantly different effects were found when excluding studies with high or unclear risk of bias or those not reporting ITT analysis. The small number of trials prevented analyses across included outcomes. In assessment of pain

intensity, the Egger's test for small study effects suggest potential publication bias ($p = 0.05$). Based on the Peter's test for binary data and the Egger's test for continuous data there was no evidence of publication bias at post-treatment or follow-up for any of the remaining outcomes. Removing studies which do not report ITT analyses was expected to produce a more conservative estimate of effect and possibly eliminate significant findings. Our ITT sensitivity analysis contradicts this expectation. One reason for this may be the lack of commonality between papers. According to one study which reported 'as treated' data, there was no significant differences between 'completer analyses' and 'intent to treat analyses' therefore the former was reported.

3.7.3 Exploratory analyses

Our exploratory analyses of intervention content point to intervention components and the circumstances of their delivery which might improve outcomes in future research. To our knowledge, this is the first review of ICT-based psychological therapies for paediatric chronic pain that considers the content and functionality of the intervention delivery platform.

3.7.4 Theoretical basis of ICT-based interventions

This review demonstrates the theoretical basis of interventions for pain management may be identified retrospectively using the Theoretical Domains Framework (Little et al., 2015). We aimed to inform current understanding of ICT-based intervention development by exploring the theoretical basis of the included studies. We were able to identify several key theoretical domains targeted by the included studies. We found that all included interventions were designed to target children's knowledge, skills (development) self-regulation abilities, memory, attention and decision processes and social support. Other key domains included in all interventions were beliefs about consequences, beliefs about capabilities and environmental contact and resources. These targets are consistent with

cognitive behavioural principles (Sharp, 2001) and with Social Cognitive/Social Learning Theory (Bandura, 1977; 2005) reported by two of the included trials.

As expected, detailed description of intervention and particularly control group content was lacking. None of the included trials appeared to target 'optimism', 'social role and identity' or to implement a theoretical domain addressing individual intent to change ('intentions'). This is interesting given that included participants are at a developmental stage where social role, perceived identity and the influence of peers can have a huge influence on a young patient's pain management experience (Eccleston, Wastell, Crombez, & Jordan, 2008; Forgeron et al., 2010; Stinson et al., 2016). Due to lack of detailed description of theoretical basis, it may be that these domains were targeted but not accurately captured in content analyses. This may also highlight an opportunity for intervention developers to target social role and identity and implementation intentions or readiness to change in future attempts to facilitate behaviour change.

Comparison of interventions which apply or do not apply to specific domains was prevented for all but three domains. There were no significant differences between those studies that included three specific domains; reinforcement, goals or emotion. This was unexpected as we assumed interventions including these domains would be associated with larger effect sizes. We found no association between the total number of theoretical domains identified and estimates of effect on pain, functioning or psychosocial outcomes. This contrasts with findings reported by Webb et al, (2010). In a meta-analysis of Internet interventions for health behaviour change, Webb and colleagues found more extensive use of theory was associated larger effects sizes. It is difficult to draw strong conclusions from these analyses due to the small number of studies. Reasons for this inverse relationship may be due to chance finding given the small number of studies included in analyses or these domains may have been applied in the included interventions but not reported. Regarding estimates of effect, associated with the application of TDF domains, the current findings are inconclusive. Further research is required to determine if interventions that target

‘reinforcement’ ‘goals’ and ‘emotion’ are insufficient and if these domains would be associated with change in estimates of effect when combined with other domains or applied in a theory congruent manner.

3.7.5 Behaviour change techniques in ICT-based interventions

This review used an established instrument (BCCTv1) to systematically code the presence of BCT’s in technology-based psychological interventions (Michie et al., 2013). This process revealed limited use of BCT’s or reference to the mechanisms of change in technology-based interventions for paediatric pain management. The presence or absence of several techniques including ‘action planning’, ‘feedback on behaviour’, ‘information about antecedents’, ‘social reward’ and ‘problem solving’ were not associated with significant differences in effectiveness. This is in contrast with the meta-analysis of Internet-based interventions for a range of health behaviours conducted by Webb et al., (2010). The authors report the use of ‘action planning’ and ‘goal setting’ had a significant impact on behaviour.

The present review found, only the BCT ‘demonstration of the behaviour’ was significant as a potential source of variability in estimates of effectiveness. Interventions which did not incorporate this technique were associated with greater estimates of effect and significant differences compared to interventions which include this strategy. This finding may be due to chance or to it may be due to how ‘demonstration of the behaviour’ was implemented. A recent review of design features used in e-health interventions found avatar-based systems were considered a poor substitute for human interaction (Morrison et al., 2012). It may be that demonstration of behaviour is more effective when observational learning is facilitated using real-person peer models rather than animation.

The BCTs associated with larger beneficial effects are also associated with differing phases of behaviour change. ‘Information about antecedents’ and ‘action planning’ might encourage initiation of adaptive coping whereas ‘feedback on behaviour’ and ‘problem solving’ are

associated with maintenance of active coping behaviour. This is consistent with the behaviour change literature suggesting the combination of post-motivation and volitional behaviour change strategies are more effective than either category alone (Sniehotta, Scholz, U., & Schwarzer, 2005; Darker, French, Eves, & Sniehotta, 2010; Dombrowski et al., 2012; Lapp & White, 2012). Further research would need to explore the relationship between engagement in ICT-based interventions and motivational and volitional BCTs.

Surprisingly, we found no relationship between the total number of BCTs identified and estimates of effect on pain, functioning or psychosocial outcomes. Again, this contrasts with Webb et al., 2010, who found the Internet-based interventions that incorporated more BCTs tended to have larger effects. Michie et al., (2009) would suggest interventions which offer fewer techniques ensure delivery and impact are more effective. Answers to this question could have important implications for intervention development. For those studies reporting disability, depression and quality of life there were no significant differences between those studies including or excluding the use of certain BCTs. One explanation for the effects of individual techniques may be related to lack of a guiding theoretical framework. Only two trials in the current review report using theory to inform intervention development and presumably the selection of intervention strategies. The reasoning behind the selection of techniques in those studies that did not report a theoretical basis is unknown. It is may be that several theoretical domains were targeted using incompatible techniques which might negatively influence evaluations of effectiveness associated with those techniques.

3.7.6 Modes of delivery in ICT-based interventions

To our knowledge this is the first review of ICT-based interventions for paediatric pain which identifies the characteristics of intervention delivery in terms of the modes of delivery that potentially contribute to intervention effectiveness. A strength of this review is the use of an established coding scheme (Webb et al., 2010) to systematically code the

usability factors applied or not applied in the included interventions. Using this scheme, we were able to identify the key design features shared by the included pain management interventions. Common modes of delivery include ‘access to an advisor’, ‘scheduled contact with an advisor’, ‘tunnelled navigational formats’ and ‘enriched information environment’. Comparative analyses of interventions with and without certain modes of delivery was possible for interventions reporting reduction in headache severity and pain-related disability. Analyses of mode of delivery in relation to reduction in headache severity revealed no significant differences between Internet-delivered and computer or telephone-based interventions. Also, no significant differences were observed between interventions that used supplementary email compared to interventions that used supplementary telephone support. A statistically significant, inverse relationship was observed between interventions that emphasised entertainment value modes of delivery including ‘quizzes’, ‘stories’ or ‘graphics’ compared to interventions that did not explicitly report the use of these modes. An explicit focus on the appearance of the therapy in terms of entertainment value was associated with significantly lower estimates of beneficial effect on headache severity compared to interventions that did not report these modes.

Analyses of mode of delivery in relation to disability was possible for interventions which feature ‘generic follow-up messages.’ The use of this mode of delivery was not associated with significantly different estimates of effect compared interventions where this mode is absent.

The limitations of the included evidence base may be one reason for these effects. Another might be that the ‘entertainment value’ modes intended to appeal to the user might have had an adverse effect. The recent review by Morrison et al (2012) suggests tailoring based on one variable and supplementary contact containing usage promotion content rather than BCTs are associated with ineffective e-Health interventions. All the included interventions rely on additional contact via email or telephone to prompt intervention usage maintain consistent record keeping.

These analyses are a first step toward identifying the factors that influence the usability of ICT-based interventions for paediatric pain. Future research might follow van Genugten et al (2016) in applying a more detailed usability taxonomy and exploring the synergistic effects of BCTs and modes of delivery on pain-related outcomes (van Genugten, Dusseldorp, Webb, & van Empelen, 2016).

3.7.7 Quality of evidence

The quality of evidence was assessed using GRADE criteria. When assessing the quality of outcomes for reduction in headache pain the quality of evidence was rated as moderate at post-treatment and low at follow-up. Treatment satisfaction was also rated as moderate in quality at post-treatment and follow-up. The remaining outcomes, including pain intensity, disability and quality of life were rated as either low or very low quality. Several studies were rated as unclear risk of bias in random sequence generation and allocation concealment domains. All but two trials (Connelly et al., 2006; Palermo et al., 2016) were rated as low risk of bias in the blinding participants and personnel domain. Only three trials used an active control condition which attempted to equalise treatment time, attention and mode of delivery (Trautmann & Kröner-Herwig, 2010; Rapoff et al., 2014; Palermo et al., 2016). Quality of evidence was downgraded largely because there are a small number of controlled trials in this field and all contain small samples or are reliant on waitlist or minimal attention control conditions, except for one trial (Palermo et al., 2016). A continued movement toward high-quality trials and new evidence is likely to change our confidence of the estimate of effect.

3.7.8 Limitations and suggestions for future research

This review was conducted in a rigorous and reproducible manner, however there are some limitations which should be noted. The search strategy was developed to extract records reported in English only and was unsuccessful in attempting to extract unpublished or close to publication

trials from grey literature. It is however unlikely that a controlled trial, reported in English was overlooked due to the high degree of current interest in this field. The scale of the current review, including just ten studies and 676 participants, limits the conclusions that can be drawn from meta-analyses, because non-significant effects may be due to low statistical power (Borenstein et al., 2009). In line with previous reviews (Stinson et al., 2009; Fisher et al., 2015), we recommend future trials aim for larger sample sizes and ensure a minimum of ten to twenty participants per arm at post-treatment and follow-up assessment. In addition to small sample sizes, methodological quality was reduced by a lack of detailed trial reporting and a reliance on passive control conditions.

Meta-analyses of primary and secondary outcomes were prevented due lack of data. These include anxiety and parental protectiveness across pain conditions and follow-up analyses of disability, depression and quality of life among children with mixed pain conditions. The benefits of ICT-delivered therapy for anxiety in children with headache and mixed pain conditions could not be determined and the effects ICT-delivered therapy for depression in children with mixed pain conditions could not be evaluated in follow-up analyses. Quantitative assessment of treatment satisfaction should be a very attainable goal in any evaluation of innovative treatment interventions. The present review was able to perform quantitative analyses for only three of ten studies that evaluate the effectiveness of novel methods of treatment delivery. As observed in previous reviews (Fisher et al., 2015; Macea et al., 2010; Stinson et al., 2009) assessment of treatment satisfaction is typically narrative. Further assessment has been prevented by the heterogeneity of instruments used to report this outcome and by the failure of individual studies to assess treatment satisfaction across trial arms. Consensus regarding optimal assessment of treatment satisfaction and assessment across trial arms would provide inform evaluations of intervention acceptability. Systematic assessment should also extend to other outcomes such as pain, disability, anxiety, depression and sleep. Future trials should facilitate systematic comparison across outcomes by including at a minimum the core outcome domains outlined in the

PedIMMPACT recommendations for the assessment of paediatric chronic pain (McGrath et al., 2008). Six of the included trials were published in after 2008 despite this, each trial omits several of the recommended core domains.

We were unable to evaluate the maintenance of treatment effects across outcomes. The number of studies included in this meta-analysis that presented follow-up data beyond a period of 3 months was limited. Two trials included in meta-analyses (McGrath et al., 1992 and Stinson et al., 2010a) presented follow-up data for a maximum period of 12 months. These studies were included in assessment of headache reduction. Follow-up analyses suggested these interventions were not associated with sustainable beneficial effects on headache severity. Future trials are needed to evaluate the long-term effects of ICT-based treatment as well as the potential mechanisms of change contributing to the maintenance of benefits.

This meta-analysis is also limited in focus and power. Where possible, future studies should explore potential mediators and moderators that may account for differences between estimates of effect. These may include participant characteristics such as age group differences, levels of pain, functioning and distress or analyses of sex in terms of completers and non-completers. These findings relate to the effects of ICT-based psychological therapy on pain related outcomes but do not explore the potential mediators (e.g., factors such as mood, self-efficacy) which might explain uptake or adherence to interventions for pain self-management (DasMahapatra et al., 2015). The included interventions are complex and not designed to evaluate the influence of isolated delivery features or single intervention factors.

In the current review, all but one of the included interventions (Palermo et al., 2016) had small sample size and all opted to include a broad age range. This is typical of complex interventions targeting multiple behaviours. However, answers to these questions would have informed current understanding of the acceptability and effectiveness of ICT-based psychological therapies in terms of child and adolescent developmental

differences, condition severity and the type of participants most likely to complete or dropout of ICT-based programmes. We echo the recommendations of previous reviews in suggesting individual trials pay greater attention to the psychological profile of participants entering treatment in terms of participant levels of pain, disability and distress (Morley et al., 2013; Fisher et al., 2015).

Lack of data prevented subgroup analyses which were planned to identify potentially effective features of ICT-based psychological interventions. We had intended to examine the relationship between estimates of effect and (i) different types of treatment delivery platforms (e.g. Internet, Smartphone, game-based), (ii) different types of psychological therapies (e.g. cognitive behavioural, acceptance-based, behavioural); (iii) different types of pain conditions e.g. musculoskeletal, abdominal, neuropathic); (iv) different categories of mode of delivery (automated, communication, supplementary); (v) contact with therapist and (vi) treatment intensity or efficiency . However, due to the limited number of trials available for inclusion and lack of detailed trial reporting we had to pool the interventions together to conduct some analyses and were prevented from performing others. While subgroup analyses did offer a closer examination of the content and methodological factors which may impact effects; the results should be interpreted with caution as the multiple statistical testing involved can increase the chance of Type I error (Munafo & Flint, 2004).

The present review took an individual rather than additive approach to content analysis. This exploration of additional theoretical domains, BCTs and modes of delivery on estimates of effect was limited by lack of data and detailed trial reporting. It may be there that there were active ingredients, such as ‘self-monitoring’, in the control arm(s) of certain studies, particularly those using a minimal attention control condition. We suggest such gaps highlight an opportunity for researchers and software developers to work more closely together during intervention development. With enough evidence, future studies might follow Dusseldorp, van Genugten, van Buuren, Verheijden, & van Empelen (2014) or van

Genugten, Dusseldorp, Webb and Empelen (2016) in exploring the additive influences of evidence-based or theory congruent clusters of BCTs on estimates of effect. This approach may be extended to analyses of modes of delivery and theoretical basis. It is possible that estimates of beneficial effect are shaped by the combination of techniques used rather than by larger numbers of BCTs. Intervention developers may benefit from greater focus on theory congruent, combinations of techniques rather than trying to incorporate a large number. The current findings should be interpreted with caution.

The findings from this review suggest that future studies should investigate alternative psychological therapies, modes of delivery and types of pain conditions in controlled trials using recommended outcomes and long-term assessment across trial arms. Intervention development should also be informed by existing evidence-based guidelines for best practice and by end-user perspectives. Interventions in this field should incorporate a higher standard of trial reporting and evaluation of the active ingredients in pain management interventions. Established intervention development frameworks may be applied and used to guide intervention development and exploratory analyses. Although these findings are tentative, the theoretical domains framework, behaviour change taxonomy and usability coding scheme employed have allowed for more accurate mapping of potential sources of variability in intervention effectiveness. These findings may help researchers and clinicians identify the circumstances in which different type of interventions are most likely to be effective.

3.7.9 Conclusions

This systematic review revealed tentative support for the effectiveness of this modality as a means of treatment delivery. While ICT-based psychological therapies were associated with significant improvement in reducing headache severity after treatment there is little evidence to support the use of ICT-based psychological therapies as a primary treatment for pain related disability, depression or to improve

quality of life. Information and communication technology appear to be an acceptable form of treatment delivery for this population. Due to the lack of detailed description of theoretical basis or intervention content it was previously unknown if the included interventions would consist of common set of ingredients. However, despite the limitations cited, commonly targeted theoretical constructs, strategies and techniques were identified. This provides valuable information that might inform future research and intervention development. In summary, the evidence analysed was deemed sufficiently promising to warrant further research.

Chapter 4 The lived experience of paediatric chronic pain: A participative study and theoretical analysis of coping strategies (Study 2).

4.1 Chapter overview

This chapter presents the qualitative, primary study conducted as part of the need's assessment outlined in Chapter 2. This qualitative study was designed to explore the experiences of paediatric pain management from the perspective of the children and parents coping with chronic pain. This chapter first outlines the rationale for this study. This is followed by a presentation of methods and results. Finally, the chapter closes with a discussion of qualitative findings and how they are applied in intervention development. Relevant literature relating to pain self-management, theoretical methods and strategies will be used to set a context for this study.

4.1.1 Introduction

The results from Study 1 provide tentative support for the feasibility, acceptability and potential effectiveness of technology-mediated CBT-based interventions for chronic headache pain management in children and adolescents. While the aggregated result in Study 1 is promising and the class effect of CBT is relatively well supported (Morley, 2011), a review of the literature reveals more mixed results for the efficacy of CBT for specific pain conditions. The findings from the preceding systematic review, combined with the initial scoping review of the literature summarised in Chapter 1, have identified key areas of focus for this research. For example, we have a limited understanding of pain management from the perspective of pre-adolescent children with chronic pain. Also, that recommended practice guidelines such as intervention as soon as non-coping occurs is typically prevented by access and resource barriers which prevent uptake of psychological support for pain management. High levels of treatment satisfaction suggest Internet-based, psychological therapy may be an

acceptable solution to these barriers. This information helped to identify an appropriate, self-management focused theory for pain self-management and guided the focus of the prospective intervention toward younger children with chronic pain and more accessible means of treatment delivery. Together, these findings suggest that further research is needed to explore how CBT-based strategies and remote modes of treatment delivery might be perceived by a younger pain population.

4.1.2 Juvenile Idiopathic Arthritis

Chronic pain is the central symptom of juvenile idiopathic arthritis (JIA), one of the most common childhood rheumatic diseases - approximately 75% of children with arthritis experience chronic pain (Prakken, Albani, & Martini, 2011). This condition, for which there is no known cause, can present in as many as seven sub-types. These include systemic, oligoarticular, polyarticular, psoriatic, enthesitis-related and undifferentiated juvenile arthritis (JA). The most commonly diagnosed types of juvenile arthritis include Polyarticular arthritis, a type of JA that involves 5 or more joints simultaneously and is usually associated with autoimmune conditions. This may be classified as polyarthritis with or without rheumatoid factor (i.e. RF-positive or RF negative polyarticular JIA). RF-positive juvenile arthritis is associated with the presence of a protein called rheumatoid factor (RF) or anti-cyclic citrullinated peptide (CCP antibody) in the blood and presents like adult rheumatoid arthritis. Extended oligoarticular juvenile arthritis is a type of arthritis that affects 5 or more joints within the first six months of onset. Enthesitis-related juvenile arthritis is a type of arthritis which presents as tenderness (enthesitis) where the bone meets a tendon, ligament or other connective tissue and accompanies the arthritis joint inflammation and RF-positive polyarticular JIA, a type of juvenile arthritis that (Tikly & Makada, 2009). Children with JIA may experience inflammation, stiffness and damage to their joints with unpredictable flare-ups causing increased pain and swelling (Rosenzweig & Nabors, 2013). Children with JIA report pain related disability that can severely limit school and social functioning (Foster,

Marshall, Myers, Dunkley, & Griffiths, 2003; Prakken et al., 2011). Approximately one third of children with JIA are likely to experience active disease which persists into adulthood (Zak & Pedersen, 2000; Foster et al., 2003). Compared to healthy peers, children and adolescents with JIA report higher levels of depression, cognitive impairment, and social and behavioural problems (Huygen, Kuis, & Sinnema, 2000; Margetić, Aukst-Margetić, Bilić, Jelušić, & Bukovac, 2005). Research indicates that children with JIA associated physical limitations may be more inclined to focus on behavioural coping strategies, and under-estimate the value of cognitive strategies for pain management (Rosenzweig & Nabors, 2013). Therefore, research that contributes to the development and improvement of effective cognitive methods of pain management for children with chronic pain is particularly warranted.

4.1.3 Pre-adolescent perspective

Paediatric pain literature reveals a dearth of studies in which children describe their experiences of coping with long term pain or their understanding of adaptive or maladaptive coping behaviour. Children learn through their experiences of pain. They develop and attach meaning to their pain based on past experiences, behavioural and environmental influences (Bandura, 1989). Therefore, it is surprising that so few studies reported on children's experiences of living with chronic pain (Sällfors, Fasth, & Hallberg, 2002; Tong, Jones, Craig, & Singh-Grewal, 2012). One exception to this is a qualitative study that used in-depth interviews to examine the experiences of coping among 22 children living with JIA (ages 6-17 years) (Sällfors et al., 2002). In that study, children and adolescents were asked: "What do you do when you are in pain?" "What do you think about when you feel pain?" and "What can you do to reduce pain?". A grounded theory methodology was applied, and several categories of coping experience were identified. Children described the instability of life with chronic pain and associated dependence, ambivalence and disturbed order. However, despite recruitment being open to younger children, the respondent sample was

comprised mostly of teenagers. This highlights a need for research from the perspective of younger children.

4.1.4 Participative person-based approach

Children with chronic pain have complex needs. To develop an intervention designed to engage young children in pain self-management, it is essential to understand their support needs from their perspective. We know little of the pain coping strategies typically employed by school-age children with chronic pain, how these strategies are perceived (i.e. utility, relevance, difficulty) or how evidence-based strategies might be prioritized and implemented in a CBT treatment approach tailored to a pre-adolescent age pain population. There are several other unanswered questions. What do school age children understand of pain self-management; what meaning do they ascribe to their pain experience; what role do they perceive they play in pain management and which therapeutic strategies are most likely to meet their needs and preferences or motivate adaptive pain behaviour. To draw conclusions about the acceptability of existing or prospective ICT-based psychological therapies for this target group we must understand their subjective experience of chronic pain management and thereby the determinants of their pain behaviour. Only then can we select and evaluate core components (e.g. CBT strategies, BCTs and theoretical basis) that might be used to target these influences (Bartholomew et al., 1998).

Several methodologies may be employed as a means of engaging children in a dialogue about their beliefs and understanding of pain self-management. Some studies involving pre-adolescent children have used write and draw, photo-voice and other image-based methods (Darbyshire, MacDougall, & Schiller, 2005; Nic Gabhainn & Sixsmith, 2006; Nic Gabhainn et al., 2007). These approaches are considered to be developmentally appropriate and offer benefits for both the researcher and participant (Darbyshire et al., 2005; Olufisayo John-Akinola, Gavin, O'Higgins, & Nic Gabhainn, 2013). A common criticism of these methods is that they involve the interpretation of the child's voice from the worldview and experience of the researcher. One alternative is the

participative research process approach developed by Sixsmith et al. (2007). This methodology allows the child to retain power in the participant researcher relationship and to contribute in all stages of the research process from data generation through to analysis and interpretation (Nic Gabhainn & Sixsmith, 2006; Nic Gabhainn et al., 2007; Olufisayo John-Akinola et al., 2013).

4.1.5 Cognitive behavioural theory

Cognitive behavioural theory posits that psychosocial factors and specifically cognitions are strongly influential on the pain experiences. Research has shown that pain beliefs and appraisals relating to pain control (having more/less control), disability (functional impaired), harm (signified damage), pain catastrophizing and self-efficacy predicted therapeutic change following cognitive behavioural therapy (CBT) for chronic pain in adults (Turner, Holtzman, & Mancl, 2007). Regression analyses have further revealed that pain beliefs are inversely related to the pain experience in children with JIA after controlling for age, sex, disease duration and activity (Thastum, Herlin & Zachariae, 2005) and that pain beliefs and coping can predict up to 29% of the variance in pain severity scores. This suggests that interventions targeting maladaptive pain beliefs and coping strategies may be more effective at reducing pain in children with JIA (Thastum et al., 2005).

CBT interventions designed to improve rheumatic pain self-management include pain education, relaxation, graded activities and cognitive-behavioural skills including attention management, thought challenging, problem solving and communication (Iversen, Hammond, & Betteridge, 2010). Efficacy evaluations of CBT as a treatment for JIA-associated chronic pain offer mixed results. Stinson et al., (2010a) conducted a pilot RCT that evaluated the efficacy of CBT-based, online self-management support for adolescents with JIA. The authors observed a significant improvement in pain scores but not in pain-related disability or health-related quality of life (Stinson et al., 2010a). In contrast, Lomholt, Thastum, Christensen, Leegaard & Herlin (2015) conducted a randomized,

waitlist-controlled feasibility trial and found improvement in participant reported quality of life but no difference in pain severity or functional disability between groups (Lomholt et al., 2015).

The therapeutic focus on cognitive as well as behaviour strategies has led to questions regarding the sensitivity and appropriateness of CBT for younger children (Kinney, 1991; Kingery et al., 2006; Freeman et al., 2007). As previously stated, children at earlier stages of development and children with severe chronic pain i.e. significant interference with physical functioning show a preference for behavioural strategies. Further research is needed to determine how CBT strategies may be perceived and used by younger children.

4.1.6 Parenting behaviour

For children, sense of agency and response to treatment may be further influenced by care-givers who have a deciding input on the treatment approach adopted. Chapter 1 briefly outlined the negative impact of over-protective parenting behaviour on adolescents with chronic pain. It may be that the impact of parenting behaviour is experienced to a greater degree among adolescents because this developmental stage is associated with a striving for independence and autonomy (Spear & Kulbok, 2004; Van Petegem, Beyers, Vansteenkiste, & Soenens, 2012). Research focusing on parental influence as a determinant of coping behaviour in response to chronic pain tends to involve samples with a wide age range therefore, we cannot compare the impact of parenting behaviour on school age and adolescent children (Claar, Simons, & Logan, 2008; Claar, Guite, Kaczynski, & Logan, 2010; Logan, Simons, & Carpino, 2012). Given the age of the proposed sample and the impact of parenting behaviour on the coping response of their children, parental involvement in treatment programmes is indicated (Palermo & Eccleston, 2009). However, the optimal nature and extent of parental involvement for younger children with chronic pain is unknown.

4.1.7 Intervention development

As stated in Chapter 2, guidelines for the development of complex interventions recommend the application of theory to facilitate understanding of the hypothesized mechanism of behaviour change (Craig et al., 2008; 2013). The needs assessment undertaken as part of this research has highlighted Bandura's (1996) social cognitive theory as a potentially useful framework to guide the development of an intervention for chronic pain management and to explain the insights offered by participants. The theoretical constructs underpinning many self-management interventions are drawn from or may be linked to Social Learning Theory (Bandura, 1977) or its successor, Social Cognitive Theory, with self-efficacy as a central component (Bandura, 1996; 2005). Bandura's (1989a; 1996). SCT is one of the three most common behaviour change theories applied in the management of chronic health conditions (Painter, Borba, Hynes, Mays, & Glanz, 2008). Explaining pre-adolescent pain behaviour in terms of SCT constructs is expected to aid the selection of intervention components most likely to engage children in a pain self-management programme and influence the desired behaviour change. As such, it may be a useful approach to understand chronic pain self-management and inform the development of novel interventions based on CBT principles

4.1.8 Purpose

This study was designed to answer research question 2: *What are the experiences, barriers and facilitators to paediatric chronic pain self-management from the perspective of the child with chronic pain and their parents or care-givers?*

The overall aim of this study was to increase our understanding of the lived experience of chronic pain management from the perspective of pre-adolescent children with chronic pain and of the factors that influence children's participation in a pain self-management. Specifically, this exploratory study aimed to (i) explore the barriers (support needs) and

facilitators (coping preferences) of adaptive pain management from the perspective of pre-adolescent and their parents and (ii) describe these needs barriers and facilitators at a theoretical level to inform further intervention development. This was achieved, first by giving children with JIA-related chronic pain an opportunity to voice their opinions in relation to their experiences of pain management, their support needs and treatment preferences. Second, by retrospectively describing these experiences and opinions at a theoretical level in a way that informs further intervention development. Social Cognitive Theory is explored as an appropriate theoretical basis with which to understand paediatric pain management.

4.2 Methods

4.2.1 Theoretical frameworks

The participative research process approach (PRP) is consistent with the participative research paradigm and person-centred ethos of this research. This underlying philosophy places emphasis on the informants' account of a phenomenon and the value of exploring the 'lived experience' of study participants (Gibbs et al., 2018). This respect for children's agency is the embodiment of Article 12 of the UN convention on the Rights of the Child (1989) and the Irish National Children's Strategy (2000) which state that the best interests of the child must be the primary consideration and the child's views must be given due weight in accordance with the age and maturity of the child (Hammarberg, 1990). Each of these reports emphasise the imperative of respecting a child's right 'to say what they think about matters that affect them and to have those views taken seriously' (Hammarberg, 1990) and of attending to children's accounts of their health problem, ensuring children have a direct input in health research (Prout & James, 1997). This ethos is consistent with Bandura's (1989) account of the child as an active participant and source of meaning and knowledge (Bandura, 1989).

4.2.2 Design

This research was conducted in line with the updated Medical

Research Council guidelines (2008) and Intervention Mapping protocol which guided intervention development (Bartholomew et al., 1998). This research adheres to the consolidated criteria for reporting qualitative research (COREQ) checklist (Appendix 10).

Initially this study design involved a series of one to one semi-structured interviews with children and parents. However, after three interviews with parent-child dyads it became apparent participants were uncomfortable and not benefiting from their contribution to this research process. Despite every effort to ensure participants would have a positive experience, the author observed that the structure of the interview process did not allow the children involved either the time or opportunity to express themselves as they naturally would. As this research is person-centred and concerned with giving a voice to a relatively neglected sub-group of the pain population, the author decided to use a developmentally appropriate and empowering participant research process outlined below.

A three-stage, participative protocol was employed. Each stage involved separate and interactive child and parent participation. Children and parents contributed in separate groups of children and parents toward each phase of the participative research process. Stage 1 involved the generation of data in response to questions; Stage 2 involved the categorisation of this data based on prevalence or importance and Stage 3 involved the arrangement and presentation of developed categories in order of importance for adaptive pain management. In the present study, each of the first three stages was undertaken by the same group of children and parents.

In line with the PRP approach employed by Daniels et al., (2014), it was the intention of the researchers to ensure the qualitative data provided by the children was kept as close as possible to its original form. A central tenet of the PRP process is that the informants' voice is unfiltered by the interpretation of the researchers. Therefore, only simple analysis was conducted for presentation purposes. The data generated by children and parents was largely derived from anonymous cards and schematic maps, therefore it was decided quotes would be labelled at a group rather than individual level. It was the intention of the researchers to avoid making

assumptions about the data provided by the children. The quotes cited in this study were selected based on their saliency and this was decided by children and parents using schematic maps. Saliency refers to the importance attributed to an idea, response or category. Children and parents separately decided on the most important categories and schematically mapped this data in order of importance. In the present study, salient responses were those identified most and in some instances least frequently.

4.2.3 Participants

Pre-adolescent children (aged 5-12 years) with chronic pain and their parents were asked to separately take part in participative research activities designed to identify their support needs and preferences. Participants were considered eligible if they had reported having experienced chronic pain for a period of three months or more.

4.2.4 Recruitment and Consent

The participants were recruited from the community via press release and social media (see Appendix 11). Recruitment and data collection took place in two areas. The first child and parent group sessions were held as part of a Family Fun day organised by a juvenile arthritis support network (Arthritis Ireland), the second location was on campus in two separate rooms provided by the School of Psychology. Working with Arthritis Ireland parents were asked via email if they would be interested in attending a voluntary workshop as part of their Family Fun Day. The circular email contained participant information sheet for parents and children outlining the purpose and design of the workshops. Prior to and on the day of the event participant information sheets, parent consent forms and children assent forms were distributed by researchers to parents who had expressed an interest in taking part (see Appendix 12 and 13). Consent was obtained first from parents and subsequently assent was obtained from children, at the beginning of each group session. Initially the response was very high with 15 parent-child dyads agreeing to take part. However, ten children withdrew themselves from the workshop when a conflicting event

was mistakenly scheduled at the same time (opening of an obstacle course). None of the parents withdrew from the session. This group are referred to as Child Group 1 (CG1) and Parent Group 1 (PG1).

The second group sessions were held on campus at the School of Psychology at NUI Galway. Working with a second parent support network (Irish Children's Arthritis Network; iCAN) for children with various types of juvenile arthritis, a similar recruitment process followed. Those parents who were interested in taking part either contacted the research team directly or notified the network coordinator of their interest. Several members travelled together to attend the workshop. Upon arrival participants were given time to look over the information sheets. Parental consent forms and children assent forms were distributed, and consent was obtained first from parents and subsequently from children. No parent or child withdrew or refused to participate. This group are referred to as Child Group 2 (CG2) and Parent Group 2 (PG2).

4.2.5 Procedure

To structure the participative group sessions, a series of primary questions and probing questions were developed. The questions outlined below, were developed based the common categories identified in a preceding qualitative literature review and from previous participative research studies (O'Higgins et al., 2013). Questions were open-ended and deliberately phrased in an upbeat, positive manner to encourage high energy during groups session. In line with a Social Cognitive Theory (Bandura, 1996), the questions asked were intended to explore the personal, behavioural and environmental, process and contextual factors that influence: (a) child and parent experiences of coping with paediatric chronic pain; (b) their use of coping strategies in daily life; (c) their perceptions of existing chronic pain support and (d) the circumstances in which self-management is perceived as feasible.

Each group session began with an introduction to the study, the purpose of the research, what the workshop would involve, what could be expected from participation and being informed that anyone could stop their

participation at any stage if they so desired i.e. the voluntary nature of their participation. This included a briefing on the confidentiality of the information shared and the phases to be completed in each group session. Data were collected and analysed for parents and children concurrently and in separate group sessions. Each group session was facilitated by an experienced researcher and research assistant who as much as possible, refrained from influencing participant's decisions. Each group session consisted of three phases. Each sequential stage involved the same child and parent group working separately through stages of the process. The group sessions with both groups lasted approximately 40 minutes. Procedure for data collection and analysis are outlined below (Table 4.1).

Table 4.1
Summary of process

Phase	Activity
Preliminary	Recruit, distribute information, secure consent/assent
Phase I	Brainstorm support needs and coping/treatment preferences
Phase II	Group cards
Phase III	Feedback on categories including what is missing and discuss relationships between categories

Phase 1: Data generation. All participants were asked to give their responses to two of the following three questions which were designed to elicit their views on potential barriers and facilitators (also described as support needs and coping preferences for ease of understanding) of adaptive coping behaviour:

Q1: *“What things do you like to do when you are in pain?”*

Q2: *“What things can you (your child) not do when you (your child) are in pain?”*

Q3: *“If it were your job to make life better for children with pain what would you do?”*

Responses were explored using the following probes:

- *“Why do you/we need to do that”?*

- *“How do you/we do that”?*
- *“What support do you/we need to do that”?*

Participants were encouraged to contribute as many ideas and responses as they want. To facilitate this process all participants were provided with rectangular pieces of coloured card and encouraged to record one idea or response per card.

Phase 2: Data analysis. In Phase 2, participants were asked to categorise their responses by playing a version of the ‘Snap’ card game. All the response cards were first collected and dealt out by the youngest member of the group, face up, to each participant so everyone had a pile of written responses to categorise. The youngest member of the group read out the first response on the table face up, and other group members then reviewed their own piles for a response they believed would match the one on the table. If a response was found to be the same or similar it was placed on top to form a category. In this way, similar responses were placed together, and different categories emerged until all the cards were used. Participants were then asked to verify they were satisfied with their choice of categories. As they decided on descriptive labels for each category and its contents were reviewed: in some cases, the pile of ideas was divided into new categories or combined with others.

Phase 3: Data presentation. All participants were involved in organizing their response categories in a schematic map of the idea(s) or issue(s) most important or most challenging for them. These issues were placed in the first layer of the map. As a group, participants then populated the outer layers of the map with their ideas on how to address the central issue. Descriptive labels for each category and its contents were again reviewed and, in some cases, removed, divided into new categories or combined with others. In addition to written responses and the creation of schematic maps, all sessions were audio-taped and transcribed verbatim.

4.2.6 Analysis

Inductive and deductive approaches to analyses were carried out by one researcher (A.T.) and verified by a second researcher (SOH). Inductive analysis involved the categorisation and interpretation of emergent data using a participative research process (PRP) methodology (Sixsmith et al., 2007). Categorisation using the format of a card game was undertaken by the participants in a manner consistent with saliency analysis whereby codes (ideas or responses) are organised into categories (themes) based on similarity, importance or how frequently they recur (Buetow, 2010). Saliency analysis ensures ideas and responses which are non-recurrent but potentially important are revealed in the analysis. The PRP approach was selected because it supports the generation, analysis and interpretation of data from the perspective of the participant without *a priori* assumptions or the filtered interpretation of the researcher. This approach is optimal in the context of this study as the views of pre-adolescent children on their experience of pain self-management are poorly understood. All data generated through group workshops including written responses, visual and audio materials were included in the data set for analysis. This approach produced a schematic overview of the data which informed the selection of a theoretical framework to conceptualise paediatric chronic pain self-management.

Deductive analyses involved a theoretical thematic analysis of participant identified barriers and facilitators to pain self-management and use of technology-based interventions in line with Social Cognitive Theory (Bandura 1996; 2005). A coding sheet was developed based on published definitions of Social Cognitive Theory constructs and used by the authors for consistent coding (Bandura, 1986; 2005) (Appendix 14). Written, verbal and visual responses were coded by hand and mapped to the main constructs of SCT (AT). Next, all written responses, utterances including identified barriers and facilitators were collated, defined and grouped into sub-themes under each construct by the first researcher. The second researcher coded responses independently to ensure fidelity of the coding guide. All coding was discussed and agreed by the two researchers.

4.3 Results

4.3.1 Demographic results

The recruitment approach targeted children aged 5-12 years with various types of chronic pain and their caregivers. Although the inclusion criteria were designed to be inclusive the resulting sample consisted only of children diagnosed with a range of juvenile idiopathic arthritis (JIA) conditions and were of White-Irish ethnicity. The demographic characteristics of both parents and adolescents are presented in Table 4.2.

Table 4.2

Demographics of participants in participative groups and think-aloud sessions (N=32)

Participant number	Child-CG1	Child-CG2	Parent-PG1	Parent-PG2	Age	Sex (M/F)	Diagnosis	Pain Duration (years)
1	*				11	F	E-oligo	4
2	*				7	M	P-oligo	1
3	*				9	F	P-oligo	1
4	*				8	F	Poly Rf-	1
5	*				9	F	Poly Rf-	2
6		*			12	F	Poly Rf-	3
7		*			12	F	Enthesitis-related	9
8		*			11	F	E-oligo	7
9		*			11	F	Enthesitis-related	5
10		*			10	F	P-oligo	4
11		*			9	F	Poly Rf-	3
12			*		43	F		
13			*		45	F		
14			*		45	M		
15			*		46	M		
16			*		42	F		
17			*		47	M		

18	*		40	F
19	*		42	M
20	*		41	M
21	*		45	M
22	*		40	F
23	*		45	F
24	*		44	F
25	*		43	F
26	*		39	F
27		*	41	F
28		*	45	M
29		*	46	M
30		*	40	F
31		*	38	F
32		*	43	F

E-oligo: Extended-oligo- articular juvenile idiopathic arthritis; P-oligo: Poly-articular juvenile idiopathic arthritis; Poly Rf: RF-positive poly-articular juvenile idiopathic arthritis. Enthesitis-related: Enthesitis-related juvenile idiopathic arthritis

4.3.1 Aim 1: Explore barriers (support needs) and facilitators (coping preferences) for adaptive pain management from the perspective of pre-adolescent and their parents

4.3.2 Phase I. Data Generation

Table 4.3 presents a summary of the data generated by each group across each participative phase of research. All participants in each group took part in each phase of the participative research process. Written responses to Q1: “*What things do you like to do even when you (your child has) have pain?*” revealed that children (n=5) generated 26 more responses than parents despite parents (n=15) outnumbering children. A total of 99 responses were generated for question 1. Overall, 53 responses were generated for question 2: “*What things can you (your child) not do when you are in pain?*” (Q2). The quantity of written responses to Q2 were similar in number between children and parents. Overall 152 responses were generated for question 3: “*If it were your job to make life better for children with pain what would you do?*” Written responses to Q3 revealed that parents (n=21) generated 28 more responses compared to children (n=11). CG1 & 2 (n=11) provided 63 individually written answers in response to Q3. PG1 & PG2 (n=21) provided 90 individually written answers in response to Q3.

4.3.3 Phase II. Data categorisation

In Phase II each child and parent group worked separately on dividing the responses generated into categories. A total of eleven children and twenty-one parents made decisions about which answers should be categorised together and decided what labels or descriptors best applied to each category. In total the children provided 155 responses and parents provided 153 responses which outlined their experiences, barriers and facilitators. These barriers and facilitators were categorised by participant and resulted in 15 categories of barriers or support needs, 20 categories facilitators or coping preferences and 29 actions that might be taken to improve pain management among school age children (see Tables 4.3 - 4.4).

There was a lot of overlap in child and parent responses and categories.

Table 4.3

Data generation across phases

Child Group 1	Children	Parents
Phases 1	n = 5	n=15
Phase 2	n = 5	n=15
Phase 3	n = 5	n=15
Q1. What things do you like to do even when you (your child has) have pain?	63 responses	36 responses
	10 categories 1 schema	10 categories 1 schema
Q3. "If it were your job to make life better for children with pain what would you do?"	35 responses	37 responses
	9 categories 1 schema	12 categories 1 schema
Features of research participants and responses from CG2		
Child Group 2	Children	Parents
Phases 1	n=6	n=6
Phase 2	n=6	n=6
Phase 3	n=6	n=6
Q2. What things can you (your child) <u>not</u> do when you are in pain?"	29 responses	26 responses
	9 categories 1 schema	6 categories 1 schema
Q3. "If it were your job to make life better for children with pain what would you do?"	28 responses	54 responses
	9 categories 1 schema	17 categories 1 schema

In response to question 1: "What things do you like to do even when you have pain?" children (CG1) reported the things they like to do, and which help when they are in pain. These largely referred to distraction, relaxation and behavioural coping strategies (Table 4.4). The category labelled "Push yourself to do it" contained the largest number of response

cards (n = 26/63) and accounted for (41%) of child responses. Most of these responses referred to sports and various self-regulation activities. Despite only two response cards, the children agreed the category labelled “*Be with Mom*” was important for good coping. Parents offered a different perspective and identified preferences for emotion-focused coping (see Table 4.5). The category “*Comfort them*” contained the largest number of responses referring to importance of affection and reassurance for better coping.

In response to question 2: “*What things can you (your child) not do when you are in pain?*”, children (CG2) described barriers to adaptive (“good”) coping behaviour in terms of what they felt they cannot do or need the most support to do when they are in pain. Children reported the impact of pain in terms of their physical limitations and ability to fit-in with peers. (Table 4.4). The category with the largest number of responses was “*Doing things you love*” with all response referring to activities or goals that are meaningful to the children. This category was not merged with the category labelled “*Play sports*” because the children drew a distinction between distracting activities they can engage in e.g. “*walk or stretch*” and the activities they really want to do e.g. “*wear nice shoes*”; “*play GAA*” (CG1). Parental responses validated and expanded on the physical and social barriers identified by the children by prioritising “*Being active*” and “*Friends*” and “*Concentration*” which referred to their concerns about the cognitive limitations associated with the children’s stage of development and long-term pain management (Table 4.5).

In response to question 3: “*If it were your job to make life better for children with pain what would you do?*”, participants’ described their understanding of adaptive pain management and how it can be improved. This positively worded question produced most of the identified facilitators of adaptive pain coping. Children prioritized meaningful activities that allow goal pursuit and normality (Table 4.4). Parents stressed a need for strategies that help to maintain emotional equilibrium (Table 4.5).

Table 4.4

Phase II: Categorised support needs and coping preferences identified by children (CG1 and CG2)

Q1: Facilitators (coping preferences):	N	Q2: Barriers (support needs):	N	Q3: Actions required	N
Relax	2	Play sports	6	Do your exercises	3
Stop yourself getting bored	11	Fit-in	5	Relax	7
Being with friends	3	Doing schoolwork	5	Have/use things that help	6
Writing	4	Doing things, you love	11	Have people who help	5
Going nice places	5	Going to school	2	Don't over-do it	7
Art	6	Sleep	2	Keep moving	4
Reading	2	Carry things	2	Go to nice places	1
Eating sweets	2	Getting out of the car	1	Medicine	1
Push yourself to do it	26	Talking about it	1	Support Groups	1
Being with Mom	2			Distract yourself	23
				Talk	2
				Think positive	2
				Nothing helps	1
	63		29		63

CG1: child group 1; CG2: child group 2; N: number of response cards

Table 4.5
Phase II: Categorised support needs and coping preferences identified by parents (PG1 and PG2)

Q1: Facilitators (coping preferences):	N	Q2: Barriers (support needs):	N	Q3: Actions required	N
Comfort them	8	Being active	7	Love them	6
Distraction	5	Friends	5	Communicate	8
Time-out	5	Concentrating	5	Fun activities	12
Pain relief	4	Sleeping	4	Relaxation techniques	13
Talk	4	Taking part in school	3	Peer to peer talk	3
Rest	3	Thinking positive	2	Include parents	3
Treats	3			Identity	2
Stay calm	2			Practical help	16
Friends	1			Distract them	15
Be near me	1			Don't focus on it	3
				Be positive	2
				Find right help	3
				Record pain	1
				Self-esteem	1
				Respect	2
				Acknowledge pain	1
	36		26		91

PG1: parent group 1; PG2: parent group 2, N: number of response cards

4.3.4 Phase III. Data analysis

In Phase III each child and parent group worked separately to present their data using schematic maps and the organised category titles generated in Phase II (see Tables 4.4 – 4.5). Participants organised their data in order of importance with the most important needs and preferences placed in the centre of the map. The structure of the schematic maps differed across groups. Some groups included just one category title per section whereas others included several category titles per section and highlighted (circled) the most important category within each section. Although labelled differently there was a lot of overlap between responses and categories generated by child and parent groups. Seven common categories

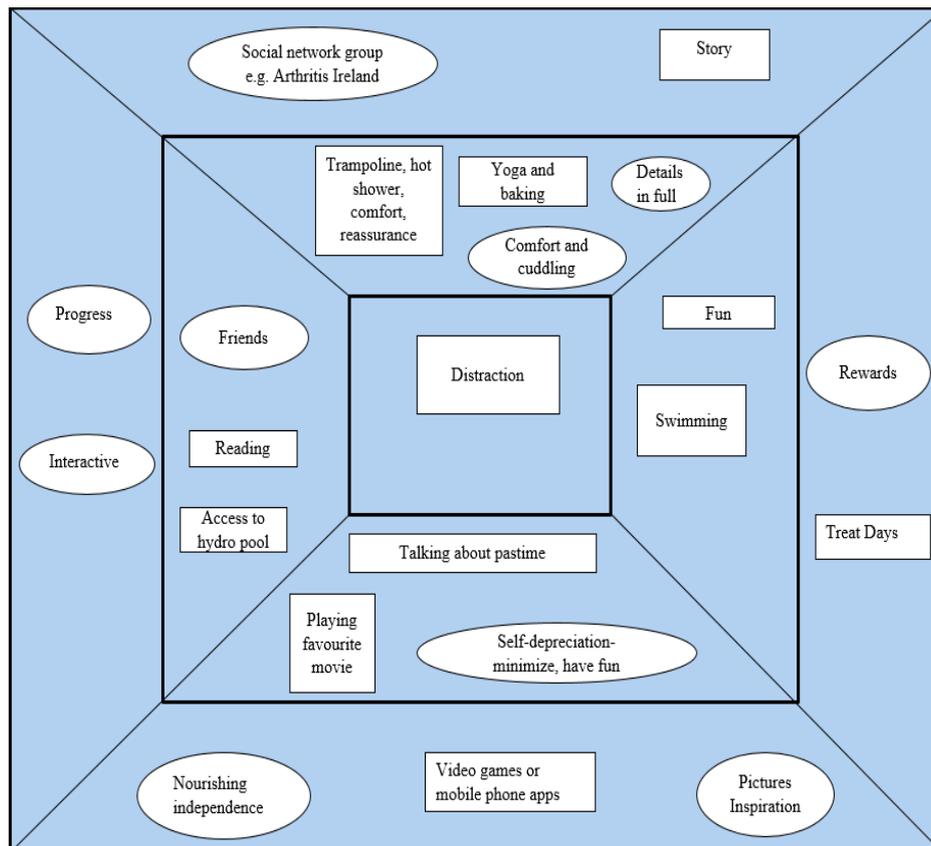


Figure 4.4 Illustration of schema, Parent Group 1 response to Question 1 (n=15)

A further four schematic maps were produced in response to Q3 and are presented in Figures 4.5 - 4.12. Several schematic maps feature multiple categories of responses, the main categories of response considered most important to participants were circled. Participants decided in phase II and III that some categories should be merged, others re-positioned, some categories were excluded, and some ideas added to the final schema (Figure 4.5-4.12). Responses referring to parties were merged with the category “going places” and the responses referring to communication issues e.g. “avoid talking about it”, “don’t focus on it” and reassurance were merged with the category “communicate”. The category labelled “treats” was merged with “fun activities” and the responses referring to “fun”, “piano”, “swimming”, “art” and “reading” were merged with the category labelled “distract yourself”. Responses including “hurling”, “when I play football I forget” and “FIFA” were merged with the category labelled “play sports”. The schema produced by both child and parent groups were organised in order of importance.

Children who participated in the study described their experience of engaging in a wide range of coping behaviours and typologies. The responses of parents largely supported these findings. The data from each child and parent group (responses and maps) were closely linked and concurred. According to each child group, the most important strategies other children should use to cope with pain are “*relaxing and thinking*” and “*don’t over-do it*”. Children identified necessary additional supports, recorded in the outer sections of their schematic map, which included: “*being alone with my own thoughts*”, “*playing various sports*”, “*relaxing with someone who listens*” “*going nice places*” “*taking medication*”, “*have hospitals closer*” and “*spacing out physical activities*” (Figures 4.5 - 4.8).

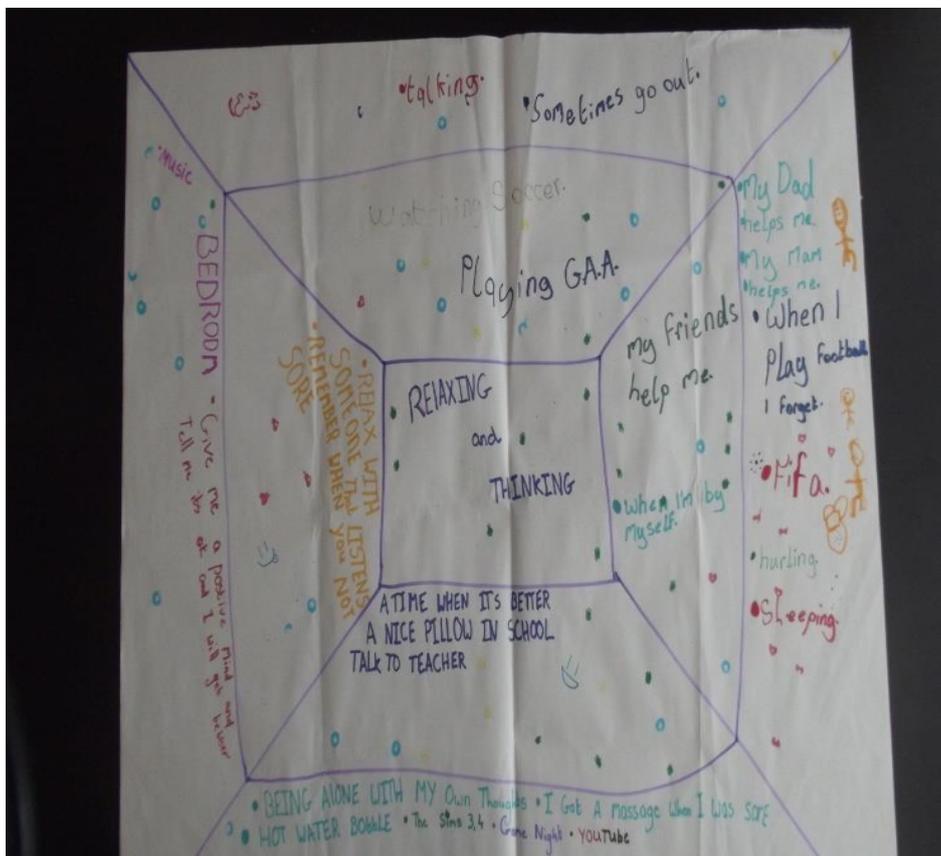


Figure 4.5 Schema, Child Group 1 response to Question 3 (n=5)

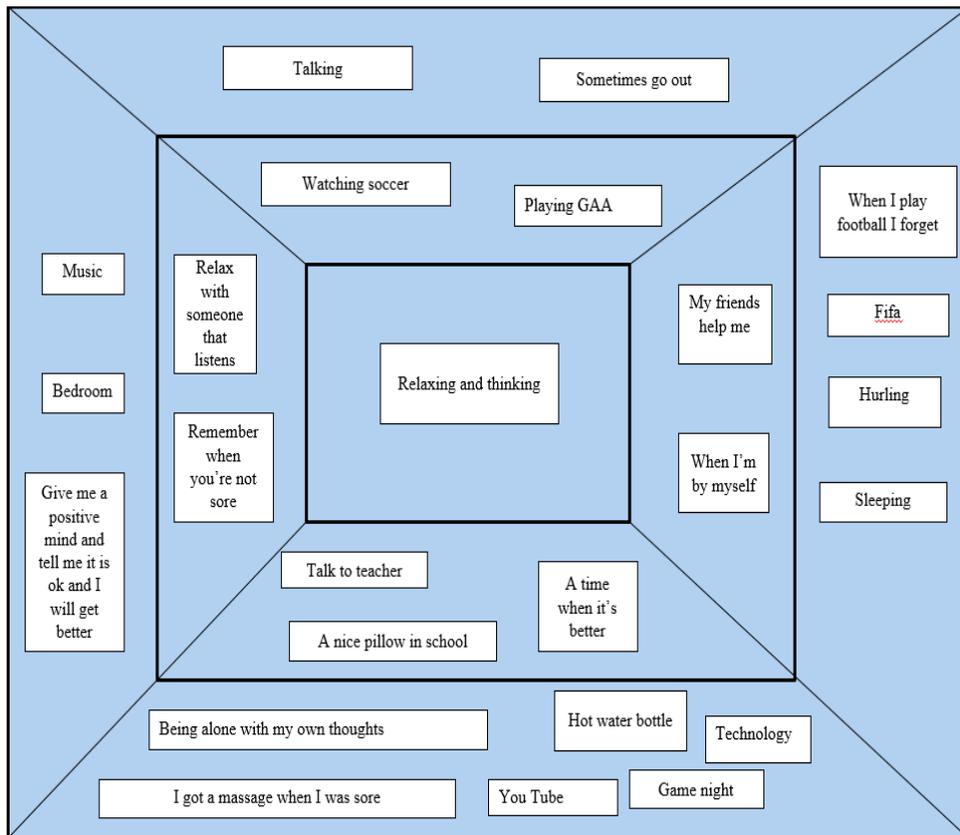


Figure 4.6 Illustration of schema, Child Group 1 response to Question 3 (n=5)

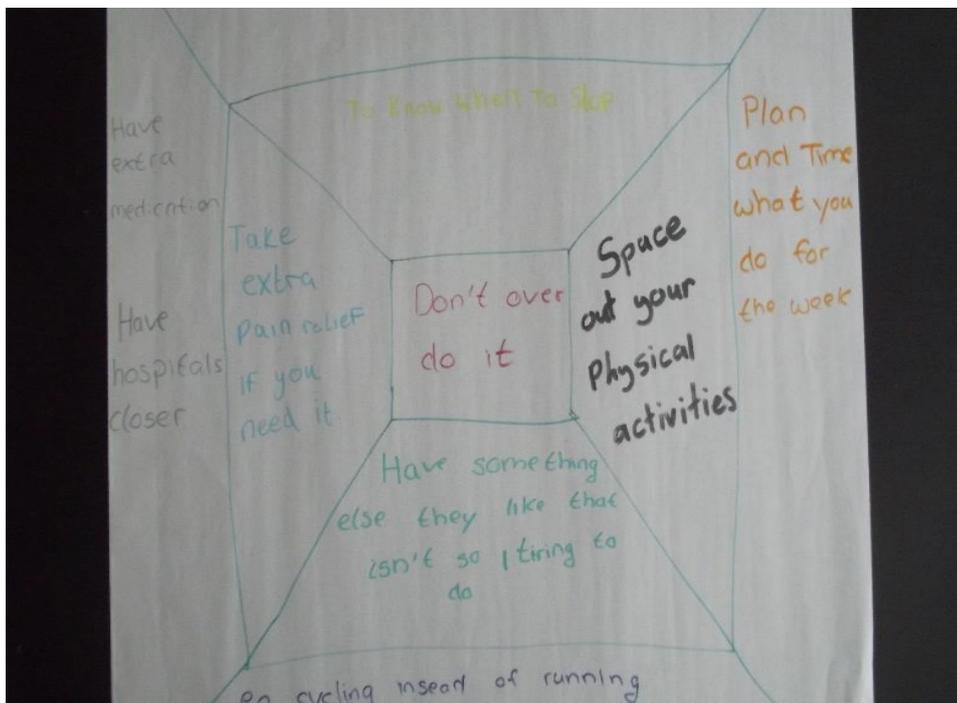


Figure 4.7 Schema, Child Group 2 response to Question 3 (n=6)

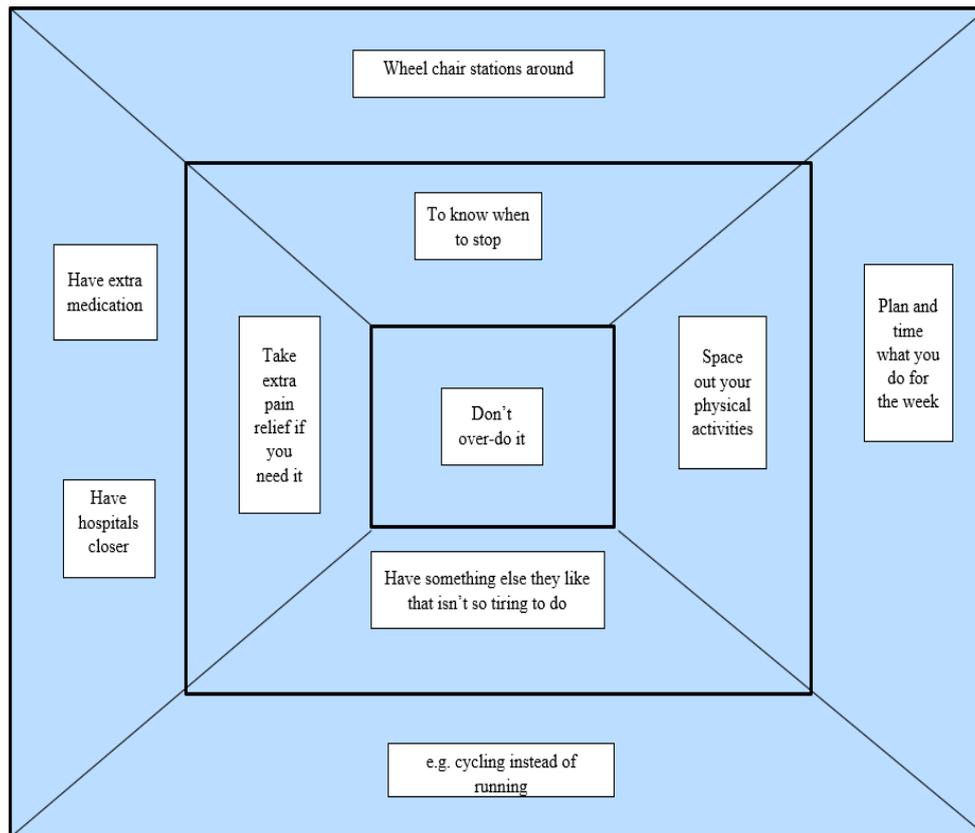


Figure 4.8 Illustration of Schema, Child Group 2 response to Question 3 (n=6)

According to each parent group, the most important strategies other parents and children should use to cope with pain are “*comfort and cuddling*”, “*show them love*” and “*find the right help*”. Parents identified necessary additional supports, recorded in the outer sections of their schematic map, which included: “*fun activities*”, “*individualised (treatment)*”, “*access to experts and other children*”, “*involve parents*”, “*empower (children)*” and “*self-management*” (Figures 4.9 - 4.12).

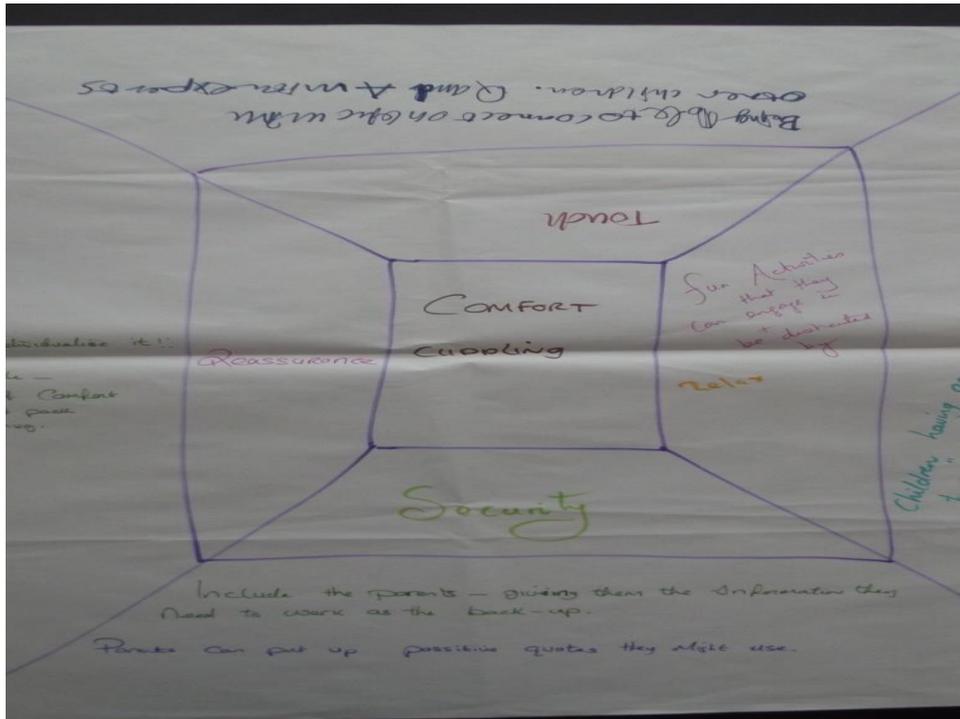


Figure 4.9 Schema, Parent Group 1 response to Question 3 (n=15)

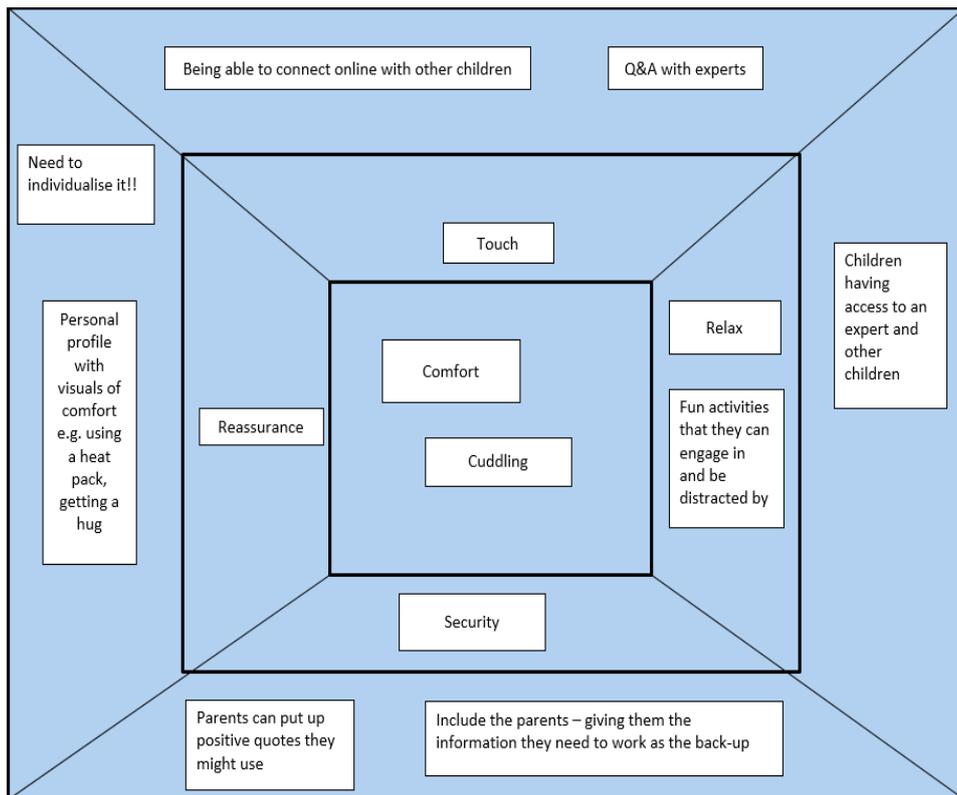


Figure 4.10 Illustration of Schema, Parent Group 1 response to Question 3 (n=15)

facilitators (preferences) of pain self-management and describe these at a theoretical level to inform further intervention development.

From these findings a list of barriers and facilitators to pain management emerged. Many of the barriers identified by participants were also suggested as potential facilitators of improved self-management. These categories were mapped to SCT constructs and barriers and facilitators were deduced (see Table 4.6). A total of five barriers and facilitators of pain self-management were identified as readily applicable in intervention development. These are presented below.

Table 4.6

Barriers and facilitators identified in PRP workshops that informed selection of intervention components

Barriers	Facilitators / SCT Constructs / Intervention components
<p>Pain Beliefs</p> <p>Children believe accessing psychological support services is a break from the norm (PRP groups). Pain beliefs, fear-avoidant behaviour or stigma associated with accessing psychological support can negatively impact coping behaviour.</p>	<p>Beliefs/outcome expectations/facilitation:</p> <p>Provide information and skills training necessary to help challenge beliefs about pain. Provide CBT-based pain education and information about the consequences (health, social and emotional) of engaging in active coping behaviour. Use a widely accessible mode of delivery. Promote the <i>Feeling Better</i> programme as a source of supplementary support to pain management in a way that is empowering for children.</p>
<p>Knowledge</p> <p>Children and parents lack knowledge and training in a variety of pain management strategies available to them (PRP groups).</p>	<p>Behavioural capability:</p> <p>Provide training in CBT-based coping skills using multiples modes of delivery (e.g. graphics, text-based instruction, audio and visual illustrations) Ensure mode of delivery is widely accessible particularly for those in rural areas or using home or portable devices.</p>
<p>Confidence in ability</p>	<p>Self-efficacy/ observational learning:</p>

Children and parents, especially those with a recent diagnosis of chronic pain lack confidence in their ability to manage pain and perform pain management skills effectively (PRP groups). Provide opportunities for accomplishment and information about how to use various psychological coping skills in practical or familiar environments and different contexts. Outline the benefits of practice and use case examples and meaningful goals as opportunities for observational learning and accomplishment.

Conflicting goals

Children (and parents) underestimated the control they have over their experience of pain (PRP groups). Children and parents want to avoid pain or pain provoking situations but also engage in meaningful activities.

Behavioural capability / Behaviour regulation / goals:

Provide pain education and information about health, emotional and social consequences of engaging in active coping. Use patient case examples and meaningful goals to challenge beliefs about pain and motivate practice. Use relevant peer models and illustrate barriers and solutions to practical problems. Promote active coping as a way of taking control (empowering).

Capability

Child stage of development will influence the impact of certain CBT strategies (e.g. difficulty remembering what skills to use, how and when (PRP groups). Lack of knowledge or motivation to use cognitive techniques is more prevalent among younger children (PRP groups).

Behavioural capability/ observational learning/positive reinforcement:

Provide information and instruction on how to perform behaviour in a graded way, where the child and parent user can build their self-efficacy for a given skill. Make the task more engaging by using interactive, audio and visual materials where possible. Present clinical content in a user-friendly way including facts, patient case-examples, creative narrative and analogy.

4.4 Theoretical analysis

Social cognitive theory (Bandura, 1989), was identified in an initial review of the evidence base as a potentially useful framework to understand chronic pain behaviour. The appropriateness of this theory is validated in the following analysis. Determining influences on pain self-management were deduced from the initial analysis of responses cards and participant identified categories carried out by children and parents. These factors were then mapped onto three larger domains: person factors, behavioural

factors and environmental factors by the research team (see Figures 4.13-4.15). This process provided a list of essential ingredients (e.g. CBT strategies, BCTs and mode(s) of delivery) that might if included, enhance the effectiveness and acceptability of the proposed intervention (Table 4.7). These factors contributed to the intervention development matrix created at the outset of this research (Appendix 2).

4.4.1 Reciprocal influence

Per the order of importance agreed by parents (PG2), environmental factors such as social support (*'find the right help'*) are a support to and are influenced by person factors such as emotional coping (*'balance emotions'*) which promote the use of coping strategies (*'self-management'*, *'communication'*) and shaped the reactions of others (*'showing them love'*) (see Figures 4.11 - 4.12). In turn, children (CG2) suggested that good coping (*'don't over-do it'*) was supported by the use of self-regulation skills ("plan and time what you do for the week") and environmental factors such as physical support ("have hospitals closer", "have wheelchair stations around") (see Figures 4.9 - 4.10). From the children's perspective (CG1) optimal self-management (*'relaxing and thinking'*) was influenced by environmental factors such as the creation of a relaxation space ("bedroom", "music" and "a nice pillow") and social support (*'have my friends with me'*, *'relax with someone that listens'*).

4.4.2 Self-efficacy

Parents reported a lack of confidence (personal self-efficacy) in their ability to correctly guide their child's practice: "*we learned from other parents*" (PG2) and "*I try to help her, but I don't really know what I'm doing*" (PG2). Lack of formal skills development training from a credible source (healthcare professional) was identified as a reinforcer of low confidence: "*we get lots of advice on meds, but no one tells us how to deal (cope) with it (pain)*" (PG1). Among children, belief in their ability to manage their pain was directly influenced by their previous experiences of disablement or lack of mastery experience. On a 'good coping day', the

children reported enduring pain and engaging in physical activity despite their discomfort: “*push yourself to do it*”; “*keep it moving*” (CG1). A ‘bad coping day’ was described as one where activity was avoided out of fear of exacerbating pain: “*Try not to move it*” (CG2, female) and “*I just can’t do it*” (CG1, female).

4.4.3 Outcome expectations

Lack of belief in the value or likelihood of benefit resulting from coping skills practice influenced child and parent motivation to use certain coping strategies. Another stated: “(psychological therapy) *she’d think we don’t believe her*” (PG1). Another stated: “*she’s afraid to do anything in case it gets worse*” (PG1). For some children, negative outcome expectations were reinforced by a perceived lack of immediate benefit resulting from the practice of certain skills that develop over time such as relaxation or mindfulness: “*there’s no point, nothing helps*” (CG1, boy, age 7) and “*I tried that once, but it doesn’t work*” (CG2, female).

4.4.4 Emotion

Emotional distress was identified by parents as one of the main barriers to adaptive coping and an important target for treatment. One parent described the experience of trying to practice a progressive muscle relaxation exercise with her daughter as a failure: “*she gets too upset – we just give up*” (PG2). Another stated: “*feeling down stops her from being active*” (PG1); “*she feels like a burden*” (PG1, mother); “*feeling down stops her looking forward to things*” (PG2, mother). To alleviate emotional distress, parent groups (PG1 & PG2) offer affection and reassurance: “*have her be with Mommy*” (PG2); “*comfort and cuddling*” (PG1), “*love them*” (PG2) and “*reassure him*” (PG1) (see Figures 4.9 - 4.12).

In contrast, fathers in the group (PG1, n = 7) saw their role as that of the “fun parent”, responsible for distracting the child from their pain whereas mothers were there as a source of emotional support. This dependent coping was cited as a source of stress by most mothers because of the conflict they experienced when trying to divide their attention among

their other children or attend to other commitments.

Interestingly this topic was largely avoided by most children. Only one child (CG1; girl, age 12) suggested cognitive strategies to manage pain: “*give me a positive mind and tell me it is ok, and I will get better*” and “*remember when you’re not sore*” (CG1, girl, aged 11) (see Figures 4.5-4.6). In line with parent observations, children in both groups identified two sources of emotional support in the form of maternal support and peer support. Only one child identified his father as a source of support.

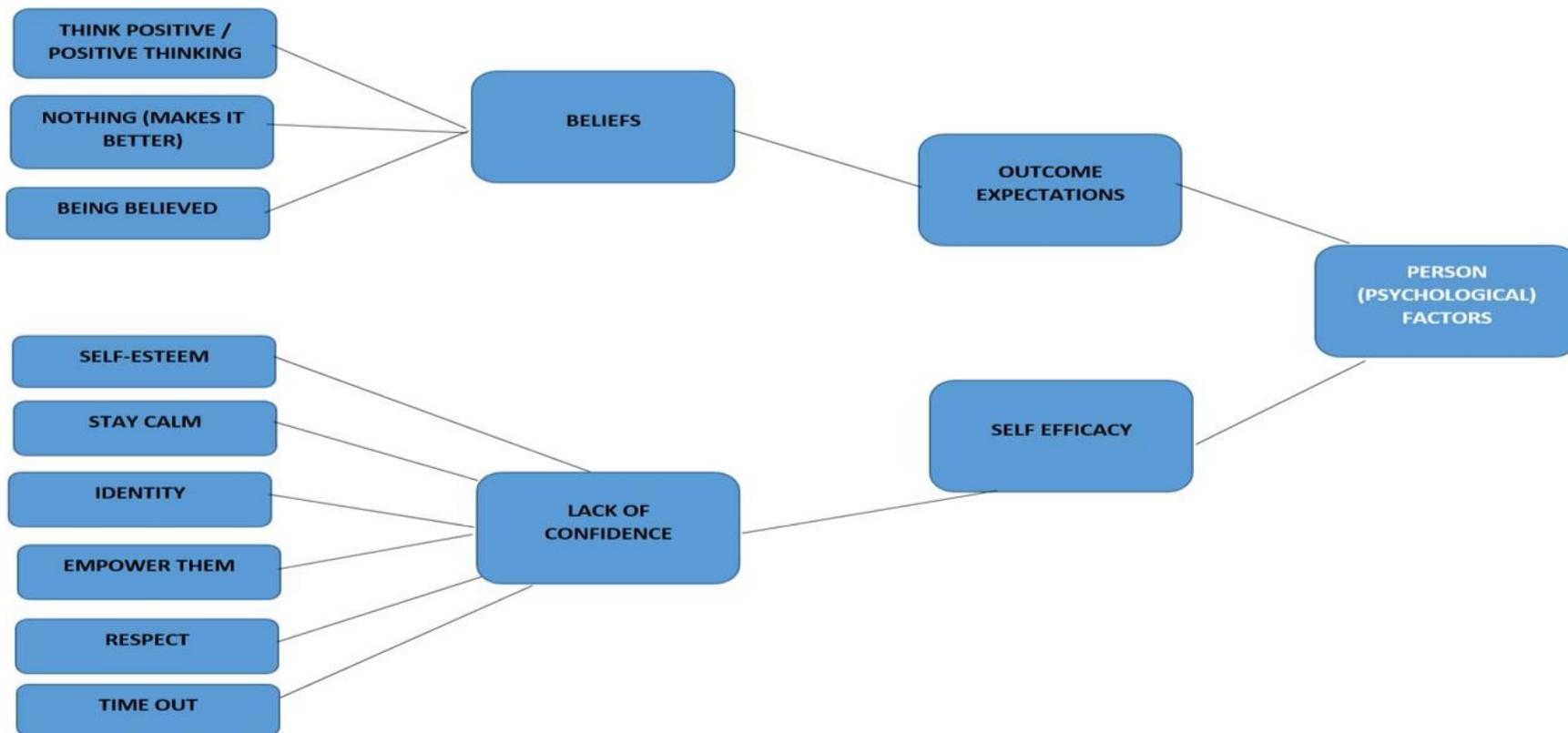


Figure 4.13 Participant response categories mapped to Social Cognitive Theory, Person factor constructs (Bandura, 1996;2005).

Behavioural factors

4.4.5 Behaviour regulation

Children and parents suggested coping strategies for pain management that were almost exclusively behavioural: “*push yourself to do it*”, “*space out your physical activities*”, “*plan and time what you do for the week*”, “*know when to stop*” (CG1, CG2) (see Figures 4.7 - 4.8). Only one child considered the influence of their thoughts on their experience of pain: “*think positively*” and “*give me a positive me and tell me it is ok, and I will get better*” (CG1). This preference for behavioural strategies was attributed to the competencies of the child participants.

4.4.6 Behavioural capability

Children report being prevented from engaging in a variety of activities due to physical limitations. Children in both groups identified activity pacing as an important approach to pain management: “*keep it moving*”, “*playing*”, “*GAA*” and “*don’t over-do it*” (CG1, CG2) (see Figures 4.1 - 4.2). In both groups, these categories contained the largest number of responses produced by children. When probed, children expanded on this theme, drawing a distinction between activities they engage in to distract themselves “*stop yourself getting bored*” and activities you engage in because they are: “*things you love doing*”.

Cognitive and emotional barriers to optimal pain management were identified as significant by parents (PG2). One parent noted her daughter would have difficulty complying with treatment and would need simple, regular instruction: “*she would need reminders*”, “*reminders of progress*” and “*details in full*” (PG2). In terms of capability, it may be that confidence rather than knowledge is the true obstacle. Despite claiming a lack of knowledge of psychological coping strategies, parent (PG2) responses from Phase I and category selection from Phase II and III suggest they used coping strategies consistent with the practice of positive reinforcement (“*encourage*” and “*reassure them*”), incentive motivation (“*rewards*”, “*treats*”) and observational learning (“*lead by example*”, “*give examples*”).

Individual coping behaviour patterns change over the course of

development in line with patient age and changing demands of life. As one child commented: *“that might have helped me years ago”* (CG2). A parent also responded: *“that would’ve been useful early on but we’re too far down the road now”* (PG2). Participant attitudes toward pain management seemed to be influenced by the duration of the pain condition and the level of frustration experienced on the pain management journey. One parent described this experience: *“at the very beginning I felt, you know you go through all the why me’s and the crying for a couple of years and then you come out of it and you get a bit stronger and you start dealing with it then”* (PG2). Children and parents suggested tailoring treatment to the individuals as essential to treatment success: *“make it (treatment) about us, not just arthritis”* (CG1); *“I’d like being able to do it myself”* (CG2), *“make it interactive”*, *“inspirational pictures”* (CG1 and CG2) and *“individualise it”*; *“personal profile”* (PG1). Participant feedback indicates a self-management support programme would be better received if it is tailored to their child’s needs and preferences.

4.4.7 Goals

Children experienced conflicting desires to avoid (e.g. *“try not to move it”*) and engage in activity (e.g. *“do your exercises your physio gave you”*); (CG2). For older children, particularly children who experienced pain for several years, coping behaviour was influenced by goal pursuit such as wanting to *“be with my friends”*, or *“go on school tours”* and goal adjustment: *“have something else you like to do that isn’t so tiring”* and *“mine can’t go to school due to pain”* (PG1) and *“take part in school activities”* (PG2). Being incapable of meaningful activities was particularly frustrating. One child describes her experience of pain: *“it stops you doing the things you love”* (CG1). Another child commented: *“a good coping day is one where we can be normal”* (CG1). For children in both groups, being normal means being capable of physically *“being able to keep up with my friends”* (CG1) and therefore *“fitting-in”* (CG1) (Figures 1-4). Parents in both groups validated this theme: *“all she wants is to be like her friends”* (PG2) and *“she’ll try anything if it means she can be like her friends”* (PG2).

Most parents acknowledged disablement as a source of stress for the child and family. Children and parents identified goal pursuit and meaningful activities to address this barrier: “*have things you love doing*” (CG2) and “*make them feel part of small activities*” (PG1). Thus, meaningful activities and behavioural goals were identified as strong motivator of engagement in pain self-management and a potential target for treatment.

4.4.8 Coping Habits

The presence of entrenched coping habits formed through trial and error learning with little formal pain education or coping skills training was cited as a significant barrier to behaviour change. Regarding parental protective behaviour and dependent coping, parents commented: “*I try not to do everything for her but it’s hard not to*” (PG2) and “*she’s like a sticking plaster to me*” (PG1). Due to the duration of practice, several parents felt changing these habits would be a major obstacle. While these categories of participant responses had the least number of recurring responses, all parents related to the experience of confusion when trying to follow pain management advice. Participants were unsure if they should “*avoid talking*”, “*talk about general stuff*” or “*acknowledge pain*” and most parents felt reluctant to draw attention to the pain and upset emotional equilibrium.

4.4.9 Incentive motivation

All parents reported relying on special treats and “*meaningful distractions*” to alleviate the emotional distress and other consequences of chronic pain. In response to the question, “What helps when your child is in pain?” parents responded: “*when my child is in pain-do her nails*”, “*make them the centre of attention*”, “*treats*”, “*distract them*”, and “*being there and encouraging your child*”. One parent requested that future interventions for pain management incorporate mindfulness (“*acceptance*”, “*mindfulness*”). This is consistent with child preferences for incentives such as “*treat days*” and “*going nice places*” to break up the monotony of coping with recurrent pain. All children (CG1 & CG2) and parents (PG1 & PG2) identified

incentives, tangible positive reinforcement and rewards for behaviour as essential to alleviate psychological distress and promote “good” coping. Frequently reported facilitators included “friends”, “fun”, “rewards” “fun activities” and “treat days” as typical facilitators (Figures 1-4). The subcategories containing the most frequently recurring responses was labelled “distract them” and “treats”.

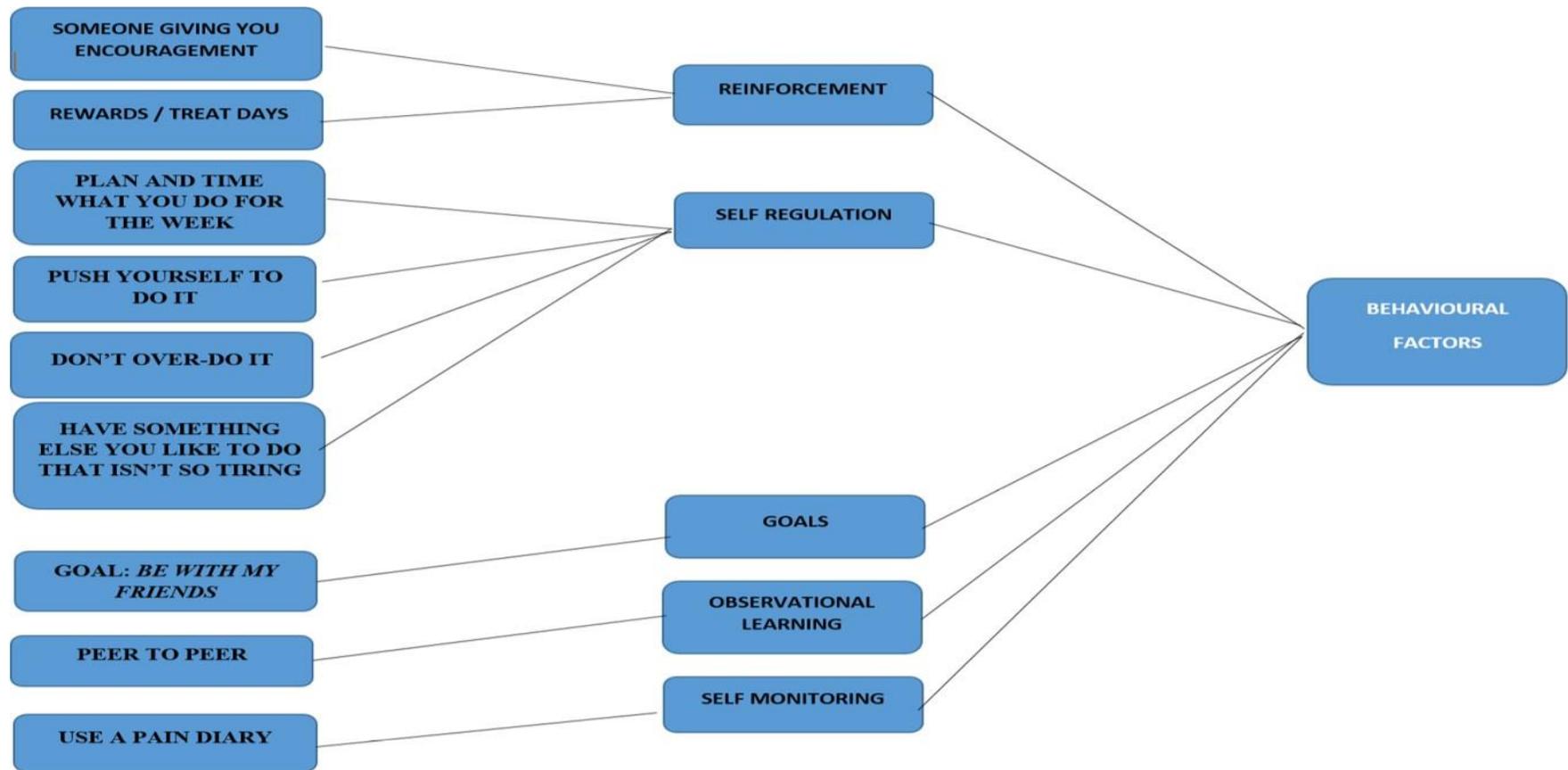


Figure 4.14 Participant response categories mapped to Social Cognitive Theory, Behavioural factor constructs (Bandura, 1996;2005).

Environmental factors

4.4.10 Social support

Among parent groups there was consensus on the two most important targets for treatment, emotional distress and environmental support (physical and social). Consequently, they produced more than double the number of responses referring to emotional and social functioning as those produced by children. Parent commented on the need to “gather friends around them” and provide “peer to peer” interaction (PG1 & PG2). Parents in both groups observed that family support networks such as the Irish Children’s Arthritis Network and Arthritis Ireland were important sources of social support and of opportunities for learning: “we came here (Arthritis Ireland, Family Fun Day) and she sees that other children need to go out to the car to rest, she doesn’t feel so bad about it” (PG1). Support group involvement served to provide advice, share information and minimise isolation, “because you realize you’re not the only one” (PG2). Parents describe active coping outside the home as more difficult to implement because their children preferred to be treated like their healthy peers and the practice of overt or obvious coping strategies had the effect of making the child feel singled out or different.

Children report social support as both a barrier and facilitator of active self-management. For most, support from peers was welcomed and essential to good coping: “my friends help me” (CG2) and “have my friends come over” (CG1) (see Figure 4.1-4.2). Others preferred to escape their diagnosis through friendship with peers who weren’t aware of it: “she doesn’t like her friends to know” (PG1). Membership to a support group was encouraged by most children: “have more support groups”; “have more groups like iCAN (Irish Children’s Arthritis Network) (CG2). Some preferred social media groups because it afforded the opportunity to create an identity without a diagnosis: “I don’t have to be sick on my profile” (CG1).

Across parent groups, a discussion evolved around the people most important to their child’s pain management. All parents identified the persons most important to paediatric pain management as mothers, peers and physiotherapists. This agreement was reached despite parent participation

which included input from seven fathers. Participants reported the influence of family as both a barrier and facilitator of pain self-management. Barriers in the form of communication issues within the home were cited as an important target for treatment by parents. As one parent comments: *“include us- Father doesn’t want to deal with her, there is an atmosphere of fear and silence”* (CG2).

4.4.11 Social facilitation

Children and parents in all groups expressed frustration when describing their experiences in the healthcare system and when trying to navigate social settings. Children identified a lack of social provision which made independence and active coping more difficult and suggested ways to rectify this, for example: *“we need more support groups like iCAN”* (CG2); *“have hospitals closer”*, *“have extra medication”*, and have *“wheelchair stations around”* (CG2) (Figures 9-10). Parents expressed frustration at access and resource issues: *“between appointments, school and work we don’t have time”* (PG1); *“if you’re not on a waiting list, you’re driving 2 hours just to get there”* (PG2) and difficulty getting a diagnosis: *“it took years, like no one believed her”* (PG2). Many child and parent participants were deeply frustrated by the lack of structural support that needlessly make coping with recurrent pain even more difficult than it should be: *“Carrying things like a school-bag”*; *“Holding things”*; *“Not being able to go on school trips”*. Some of the solutions suggested including: *“have hospitals you have to go to closer”* and *“use laptops for books”*(CG1 & CG2).

The barriers (needs), facilitators (preferences) and actions recommended by children and parents which were readily applicable in intervention development are illustrated in Table 4.7 which outlines the determinants of pain self-management and intervention components that might address these factors.

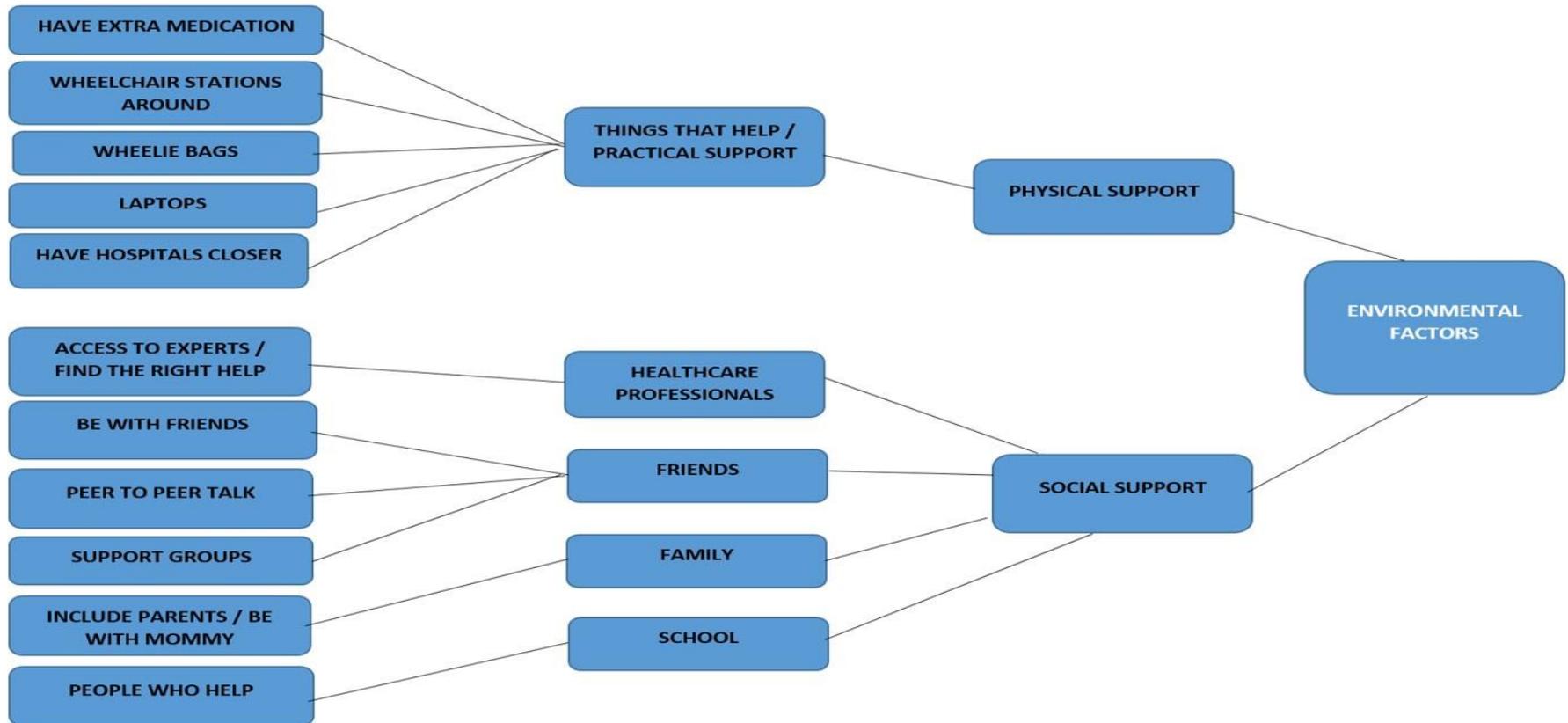


Figure 4.15 Participant response categories mapped to Social Cognitive Theory, Environmental Factor constructs (Bandura, 1996; 2005).

Table 4.7

Determinants of pain self-management and core intervention components

Determinants of self-management behavior	Needs assessment: evidence of the determinant	SCT construct	CBT-based intervention strategies	Behaviour change techniques as per behaviour change technique taxonomy v1 (Michie et al. 2013)
Pain beliefs: (cognitions-affect)	Children (and parents) under-estimate the control they have over their experience of pain: Systematic reviews of mediators, moderators, predictors of therapeutic change in CBT for chronic pain (Turner et al., 2007); Literature review, PRP groups/ Study 1 - 3).	Knowledge; Self-efficacy: To increase participants perceived competence to perform each SM behavior	Active learning; Verbal persuasion;	1.2. Problem solving, 2.2 Feedback on behaviour, 2.3. Self-monitoring of behaviour, 2.7 Feedback on outcome of behaviour, 3.1. Social support (unspecified), 3.2. Social support (practical), 3.3. Social support, (emotional), 8.1 Behavioural practice/rehearsal, 8.7 Graded tasks, 12.5 Adding objectives to the environment, 15.1. Verbal persuasion about capability
Pain catastrophizing (cognitions-affect)	Children express negative expectations about the consequences of using coping strategies. Systematic reviews of mediators, moderators, predictors of therapeutic change in CBT for chronic pain (Turner et al., 2007); Literature review, PRP groups/ Study 1 - 3).	Outcome expectations: To reduce negative expectancies (catastrophizing) about consequences of engaging in specific SM behaviours	Active learning; Verbal persuasion;	2.2 Feedback on behaviour, 2.3. Self-monitoring of behaviour, 2.7 Feedback on outcome of behaviour, 5.1 Information re health consequences of behaviour, 5.6 Information re motional consequences of behaviour, 9.1. Credible source

Table 4.7

Determinants of self-management behaviour and core intervention components (continued).

Determinants of self-management behavior	Needs assessment: evidence of the determinant	SCT construct	CBT-based intervention strategies	Behaviour change techniques as per behaviour change technique taxonomy v1 (Michie et al. 2013)
Self-efficacy (confidence in ability)	Children (and parents) lack confidence in their ability to cope with pain: Systematic reviews of mediators, moderators, predictors of therapeutic change in CBT for chronic pain (Turner et al., 2007); Literature review, PRP groups/ Study 1 - 3).	Self-efficacy: To increase participants perceived competence to perform each SM behaviour	Active learning; Verbal persuasion; Self-monitoring of behaviour;	1.2. Problem solving, 2.2 Feedback on behaviour, 2.3. Self-monitoring of behaviour, 3.1. Social support (unspecified), 3.2. Social support (practical), 3.3. Social support, (emotional), 8.1 Behavioural practice/rehearsal, 8.7 Graded tasks, 15.1. Verbal persuasion about capability
Fear	Children believe accessing psychological support services is a break from the norm (stigma). Fear-avoidant behaviour can negatively impact coping behaviour: Systematic reviews of mediators, moderators, predictors of therapeutic change in CBT for chronic pain (Turner et al., 2007).	Outcome expectations (personal factors), To reduce negative emotional responses (fear) of specific SM behaviours	Active learning; Verbal persuasion; Self-monitoring of behaviour	3.3 Social support—emotional; 5.6 Information re emotional consequences of behaviour; 11.2 Reduce negative emotions

Table 4.7

Determinants of self-management behaviour and core intervention components (continued).

Determinants of self-management behaviour	Needs assessment: evidence of the determinant	SCT construct	CBT-based intervention strategies	Behaviour change techniques as per behaviour change technique taxonomy v1 (Michie et al. 2013)
Knowledge	Child stage of development will influence the impact of certain CBT strategies (e.g. difficulty remembering what skills to use, how and when (PRP groups): PRP groups/ Study 1 - 3).	Knowledge: To increase participants knowledge for each SM behaviour	Active learning; Verbal persuasion;	2.2 Feedback on behavioural, 4.2 Information about antecedents of behaviour, 5.1 Information about the health consequences of behaviour
Skills	Children (and parents) engage in maladaptive coping skills e.g. dependent coping / over-protective parenting response: Systematic reviews of mediators, moderators, predictors of therapeutic change in CBT for chronic pain (Turner et al., 2007); Literature review, PRP groups/ Study 1 - 3).	Self-regulation, coping skills: To increase participants use of SM behaviours	Active learning; Verbal persuasion; Modelling; Feedback; Reinforcement; Guided practice; Self-monitoring of behaviour;	1.1. Goal setting (behaviour), 1.2. Problem solving, 1.3. Goal setting (outcome), 2.3. Self-monitoring of behaviour, 4.1 Instruction on how to perform the behaviour, 6.1. Demonstration of the behaviour, 8.1 Behavioural practice/rehearsal, 8.6 Generalisation of the targeted behaviour, 8.7 Graded tasks, 13.2 Framing/reframing, 12.6 Body changes

Table 4.7

Determinants of self-management behaviour and core intervention components (continued)

Determinants of self-management behaviour	Needs assessment: evidence of the determinant	SCT construct	CBT-based intervention strategies	Behaviour change techniques as per behaviour change technique taxonomy v1 (Michie et al. 2013)
Behaviour regulation	Children need guided practice and 'real-time' reminders establish adaptive coping habits: Systematic reviews of mediators, moderators, predictors of therapeutic change in CBT for chronic pain (Turner et al., 2007); Literature review, PRP groups/ Study 1 - 3); Barriers and facilitators identified in PRP workshops (Study 2-3).	Self-regulation, behavioural goals: Strategies to manage or change objectively observed / measured SM behaviour	Active learning; Verbal persuasion; Guided practice; Goal setting; Tailoring; Graded tasks; Individualisation; Self-monitoring of behaviour;	1.1. Goal setting (behaviour), 1.4 Action planning, 1.5 Review behavioural goal, 1.6 Discrepancy between current behaviour and goal, 1.7 Review outcome goal, 1.8 Behavioural contract, 2.3 Self-monitoring of behaviour;
Social support	Social influences have a huge impact on the coping response, children are particularly reliant on and value support from family and peer: Systematic reviews of mediators, moderators, predictors of therapeutic change in CBT for chronic pain (Turner et al., 2007); Literature review, PRP groups/ Study 1 - 3).	Environment - social & physical, impediments	Reinforcement; Facilitation	3.3 Social support—emotional, 5.6 Information re emotional consequences of behaviour, 11.2 Reduce negative emotions

Table 4.7

Determinants of self-management behaviour and core intervention components (continued)

Determinants of self-management behaviour	Needs assessment: evidence of the determinant	SCT construct	CBT-based intervention strategies	Behaviour change techniques as per behaviour change technique taxonomy v1 (Michie et al. 2013)
Behavioural capability / Motivation	Lack of motivation and capability to use cognitive techniques is a typical barrier particularly among younger children (PRP groups). Physical capability may impede practice, guided practice and graded coping strategies are indicated.	Self-efficacy / observational learning: To improve autonomous motivation of participants to engage in each SM behaviour	Active learning; Verbal persuasion;	1.1 Goal setting (behaviour), 1.2 Problem solving, 1.3. Goal setting (outcome), 1.4 Action planning, 1.5 Review behavioural goal, 1.6 Discrepancy between current behaviour and goal, 1.8 Behavioural contract, 3.1. Social support (unspecified), 3.2. Social support (practical), 3.3. Social support (emotional), 5.1 Information about health consequences, 6.2 Social comparison, 8.7 Graded tasks, 9.1. Credible source, 10.4 Social rewards

4.5 Discussion

Increasing the use and range of adaptive coping strategies among school age children with chronic pain has the potential to improve overall quality for life for the child and family and prevent maladaptive coping behaviour becoming entrenched. This study provides insight into the factors that influence the chronic pain management and the barriers and targets for treatment that are important to pre-adolescent children with chronic pain and their parents. The subjective experience of coping with chronic pain has to the best of our knowledge, never been explored in this age specific cohort. To our knowledge this is the first study on this topic to enable children and parents to participate actively in each phase of the research process, to develop data based on their own thoughts and experiences and to identify potential solutions.

4.5.1 Patterns of pain management

Rather than adopt one coping profile, children and parents in the present study report the practice of several coping typologies over time. In separate groups, children and parents described the practice of coping skills and typologies which range from self-reliant to avoidant, dependent and inconsistent coping approaches. These coping profiles are consistent with the coping typologies identified by Walker et al (2008). Overall the children and parents in our study expressed positive beliefs about engaging adaptive coping behaviour. This was largely attributed to parent and child involvement in a close-knit support group. However, dependent coping, specifically an over-reliance on maternal support was identified as a significant stressor. Many child participants held a fragmented view of pain management and attributed this responsibility to others. Children and parents also differed in the importance they attribute to different pain coping strategies. For children, meaningful activity and goal pursuit strongly influenced engagement in active coping behaviour. For parents, strategies that facilitate emotional equilibrium and harmony at home were most important.

Children and parents in this study report deterioration in quality of life and frustrated attempts at pain self-management which are exacerbated by a lack of access to healthcare professionals, pain management training and lack of social provision. These issues were reported to have had a negative influence self-efficacy and encouraged avoidant or dependent coping behaviour. This observation is consistent with research that identifies the negative impact of chronic pain if unaddressed or misdiagnosed (Mathews, 2011). Prolonged deterioration in physical, social and emotional functioning has been observed in longitudinal studies of the effects of waiting for treatment for adolescents with chronic pain (Palermo et al., 2019). Qualitative pain studies have not previously explored the effects of waiting for treatment on younger children. This research makes an important contribution to this understanding by demonstrating that like adults (Lynch et al., 2007), younger children (5-12 years) are negatively influenced by environmental factors that shape their pain beliefs.

4.5.2 Support needs and coping preferences

Seven common categories of responses were identified separately by both children and parents which illustrate barriers (support needs) and facilitators (coping preferences) experienced by this group. These include i) *Being active* (physical capability) (ii) *Things you love doing* (meaningful activities), (iii) *Be with Mom* (dependent coping habits), (iv) *Emotion / relief from emotional distress*, (v) *Friends and practical help* (social support / provision), (vi) *Concentration* (cognitive capability) and (vii) *Find the right help* (training). According to children and parents these factors were considered both barriers and if addressed, also facilitators of adaptive coping. We identified barriers which have been noted in previous research with pain populations such as social support and logistical barriers. We also identified sub-categories of responses which are unique to paediatric populations, these include developmental issues and parental dependence (Lebovidge, Lavigne, Donenberg, & Miller, 2003; Tong et al., 2012).

The most salient facilitator of adaptive coping identified by children was the role of behavioural goals and meaningful activities in active coping behaviour. The observation that goal pursuit is an important determinant of pain coping behaviour among adolescents is not a novel finding. Fisher and Palermo (2016) identified individual goal pursuit and child and family factors as potentially modifying variables of the pain response that have been overlooked in previous iterations of the Fear Avoidance Model of Chronic Pain (Fisher & Palermo, 2016). These findings suggest meaningful goal pursuit is equally important to younger children, who have less independence and control of their treatment approach which is largely managed by caregivers.

Participants identified the value of social support in pain self-management. Most parent and child participants reported having developed relationships that enhance their personal resources for coping with pain. This is consistent with much of the pain literature. Surprisingly, a minority of children and parents observed certain types of social support e.g. group membership can be overwhelming and potentially a negative influence by promoting pain catastrophising. Some children valued the opportunity offered by technology to create an identity that did not revolve around JIA. One parent suggested that while there are many benefits to accessing parent-led support networks, their influence can be overwhelming particularly in the early stages of pain adjustment and not necessarily beneficial if coping skills training has been inadequate. In line with this support need, coping strategies that target pain-related anxiety and depression and promote emotional balance were the most requested strategies in each parent group.

4.5.3 Extant research

Our findings are consistent with previous studies. Tong et al (2012) conducted a thematic synthesis of qualitative studies exploring children's experiences of living with JIA. In line with the current study the authors reported core themes consistent with the importance of 'fitting in' as identified by children (Tong et al., 2012). These findings are also in line with similar research conducted by Sällfors, Hallberg and Fasth (2001).

Sällfors and colleagues interviewed children with juvenile chronic arthritis (ages 6-17 years) on their experiences of coping with pain. The authors used grounded theory analysis to identify descriptive categories close to those produced in the current study. Sällfors and colleagues identified a core category labelled “controlling strategies” which corresponds to a category in the current study labelled by children as “Don’t over-do it”. Subcategories listed by the authors such as “planning and doing one’s best” mirror the subcategories identified by children in the outer layer of their schematic maps labelled “plan and time what you do” and “push yourself to do it”. Interestingly the authors observed most of the children in their study were adolescents who have grown into their chosen coping behaviour over time supporting the conclusion that early intervention is important to prevent maladaptive coping typologies becoming entrenched.

The barriers and facilitators identified in the current study are also consistent with those identified in previous studies of self-management of paediatric chronic illness from the parent perspective. Mansour et al (2000) reported parent-perceived barriers to asthma self-management for children (ages 5-12 years). In line with the present study, the most salient barriers reported were patient and family characteristics, health beliefs, social and physical environment. These findings suggest the barriers identified are still relevant.

Our study findings were contrary to the qualitative study by Fereday et al., (2009) regarding the experiences of children and young people (ages 4-16 years) living with chronic illness. The authors found chronic disease was not perceived as a barrier to participation in physical activity and children and parents held positive beliefs about fitting in with peers. Aside from differences in patient population, Fereday et al (2009) observed that differing responses may be due to the wording of the questions posed. For example, “what kinds of things does asthma stop you doing?” versus “what kinds of things do you like doing?”. To address this, the current study posed two versions of the same question: “What things do you like to do even when you have pain?” and “What things can you not do when you have pain?”. Despite both positive and negatively focused wording, the overwhelming response from children was that chronic or recurrent pain is

perceived as a barrier to participation in physical activity and is a barrier to the sense of belonging with peers. This was observed despite the generation of fewer responses to negatively worded questions compared to positively worded questions.

4.5.4 Children's ability to participate

Developmental barriers in the form of cognitive and behavioural capabilities (i.e. learning, memory, concentration) was submitted as a written response by only one parent in each group. However, this issue was identified as very important by all parent participants in group discussion. This concern is supported by evidence which shows pain has a negative effect on cognitive performance and attentional processes (Eccleston and Crombez, 1999; Moriarty, Finn & McGuire, 2011). According to Simons Elman and Borsook (2014) explicit and implicit learning processes are implicated in the maintenance of chronic pain with the former being the more difficult to modify (Flor, 2012; Simons et al., 2014). Implicit learning processes include operant conditioning and social learning (reinforcement contingencies and modelling) and should be targeted to address pain behaviour. Cognitive limitations might be addressed by targeting implicit learning using observational learning methods (vicarious learning, modelling) and consideration of the psychological processes relating to attention, retention, production and motivation as outlined in Social Cognitive Theory (Bandura, 1989a, 1996).

4.5.5 Implications for treatment and intervention development

Developmentally appropriate information and skills training e.g. easy to understand, graded and guided instruction is indicated. These findings highlight a need for need for an evidence-based intervention that can be easily adapted for younger age groups, introduced early and delivered in a user-friendly and accessible way. It is important the identified gaps between recommended and practiced pain management support are addressed, as the current findings suggest coping behaviour may be more malleable among those at an earlier stage of development and earlier stage

of the pain journey. This is in line with best practice treatment recommendations suggest support should begin as soon as non-coping occurs (McGrath and Frager, 1996). Support in the form of access to information and coping skills training earlier in this journey would be welcomed by parents and children.

Participants in this study emphasized the changing nature of self-management needs over the course of their journey from diagnosis to acceptance of their new normality. Participant feedback suggests pain self-management interventions that are tailored to the psychological profile of participants and involve pre-treatment and periodic re-assessment of self-management needs are more likely to engage and retain participants (Turk, 2004; Morley & Williams, 2015).

Our results suggest that a self-management intervention that facilitates meaningful goal pursuit and activities could engage younger children with chronic pain. Also, ICT-based therapy delivery that helps to relieve pain and associated symptoms but conflicts with desired goals will not be well received. This has important clinical implications. Treatment goals should be determined in collaboration with the child and in line with quality of life as defined by the child with chronic pain.

Pain management training i.e. skills development should be empowering for the participant, particularly where the treatment protocol is complex and managed entirely by the parent. Children might benefit from a self-management programme that teaches self-monitoring and future planning skills to facilitate the transition of pain management responsibility to the child in time. Skills development for this group should include cognitive coping strategies, and promotion of parent and child self-efficacy.

This study highlights the important role played by parents in enabling their children to participate in adaptive pain self-management. This is consistent with research that shows parental stress is associated with poorer psychological adjustment in parents and children with chronic conditions (Cousino & Hazen, 2013). The implications of this are that the involvement of parents or caregivers in a pain self-management programme

for younger children with chronic pain may be essential to its success and parent-child cooperation may be essential to promote engagement and adherence to psychological therapy for pain management.

Participant feedback indicates a widely accessible web-application could be an acceptable source of support for several reasons. Firstly, because children and parents experience barriers to effective pain self-management which include physical limitations and mobility issues, lack of social support and social provision. Access issues might be addressed by providing guided self-management training within the home rather than a central location which requires travel, additional appointments and further extraction from normal, day to day living. Children and parents could have more opportunity to engage with a self-management programme accessible from home potentially resulting in improved outcomes. Second, because issues around family cohesion and communication may be modified more easily using a home-based intervention as opposed to learning in a geographically remote location which family members find difficult to attend. Based on child and parent data, an easily accessible, self-management intervention which includes communication or problem-solving training is indicated and should include training on how to ask for help, how to talk to healthcare professionals and how to communicate when distressed.

A tailored pain management programme is indicated. There is a growing body of literature which supports the importance of personalizing and tailoring treatment to the psychological profile of the participant (Vlaeyen & Morley, 2005) and to participants preferences in terms of intervention content and design features (Morrison et al., 2012; Yardley et al., 2015; 2016). A pain management programme which is also tailored to parent's knowledge base may engage those parents who have themselves become 'experts in pain management' or established a degree of knowledge beyond basic introductory education materials. More effectively engaging parents could potentially improve overall adherence to a pain management programme.

Parent participants (PG1 & PG2) agreed social support in the form of access to experts, peer to peer interaction and integration with usual care should be a priority for future pain management interventions.

Taken together, these findings validated the selection of SCT as a guiding theoretical framework and shaped the first iteration of the *Feeling Better* prototype. The specific experiences, needs and preferences perceived as significant for children and parents, can be described using the concepts of SCT (Bandura, 1996). Thus, SCT may be useful to explain and understand the subjective experience of chronic pain management from the perspective of pre-adolescent children and their care-givers. It may be useful as a guide to intervention development and the selection of key components (i.e. optimal CBT-based strategies, behaviour change techniques and modes of delivery).

4.5.6 Strengths and limitations

This study has several strengths and limitations, most of which are inherent in qualitative and participative studies. Although the current sample size is suited to our study design, the small sample size offers only a snapshot of the lived experience of paediatric chronic pain management. Limitations associated with participative research in a group setting include the potential for bias associated with peer and adult influences and concerns that quieter children are not being heard. This was minimised by setting ground rules at the beginning of each PRP workshop and keeping the groups sizes small. Given the method and response to recruitment, self-selection bias is also likely. Also, despite efforts to avoid researcher bias which included making participants as comfortable as possible, the presence of the researcher may also have been an influence. While the process of eliciting insight was organic it was dependent on the circumstances of the time in which this research was conducted and as the current findings indicate, support needs change.

The recruitment approach used left the study open to extensive dropouts. Recruiting on a “Family Fun Day” proved difficult given the

nature and timing of the entertainment scheduled for the day. For this reason, 11 out of a potential 22 children withdrew from the PRP group workshops on the day of data collection. It may be that the children who withdrew had more positive or negative experiences with pain self-management and their input would have resulted in the identification of different barriers and facilitators. We were also unable to involve health professions in this study design due to ongoing difficulties with recruitment. This is unfortunate as input from the perspective of health professionals would have been a valuable addition to this exploratory study.

Vocabulary limitations may have influenced the responses offered by younger children. Research suggests children as young as 18 months of age have a vocabulary for pain, however the word “pain” may not emerge until much later in the child’s linguistic development (Stanford, Chambers, & Craig, 2006). The children in the present study, might have understood their pain and coping habits differently to the terms and definitions used by the researchers. To address this, we used several different terms for pain and pain management including feeling “hurts”, “sore”, “sick”, “good pain day”, “bad pain day” “good coping day”, “bad coping day” “what helps” or “what hurts”. However, the possibility remains that children’s responses were based on other “pains” and not just those associated with their chronic pain condition.

The PRP methodology allowed us to explore children and parents’ individual experiences of pain self-management using a methodology that was empowering for the participant. The methodology employed in the current study minimises errors of interpretation. The creation of schematic maps is a developmentally appropriate way of presenting an overview of the research findings (Piaget, 1962; 1964). We believe the use of schematic maps as visual aids to the Phase III interpretation process allowed the children to follow discussions more easily. The use of schematic maps may have eased the cognitive load for younger children and allowed participants the time to reflect on their past experiences and come back to certain issues or responses (van Merriënboer & Sweller, 2010). Involving children with chronic pain in every phase of this research was a success in this study

because of the value of the information provided. Collaboration with children for the purpose of this research improves the relevance of the data.

Multiple barriers to pain self-management were reported by participants however, due to the method of recruitment we were unable to evaluate differences in perceived barriers consistent with socioeconomic status, gender or geographic location. It is possible these factors could be addressed in future quantitative studies. Future studies might also consider

Thematic analysis involves the grouping of similar, recurring or connected codes. This analytic approach can mean non-recurring but potentially important codes are lost or obscured. The saliency analysis undertaken as part of the PRP approach is considered a safeguard against this limitation (Buetow, 2010). It should be noted the factors that prevent pain self-management among pre-adolescent children may differ to those identified in the current study due to the exposure of our sample parent and peer-led support networks. Future research should consider a different sample and setting to discover additional facilitators and barriers that might be used to develop an effective, tailored SMS programme.

4.5.7 Conclusion

This research contributes to our current understanding of the lived experience of paediatric chronic pain management from the perspective of pre-adolescent children coping with JIA-associated chronic pain and their parents. An objective of this study was to give children with chronic pain and their care-givers an opportunity to voice their opinions in relation to their experiences of pain management, their support needs and coping preferences. This was achieved. Children and parents identified many issues that need to be addressed to increase the use of adaptive coping strategies and uptake of psychological therapy for pain management support. Common pain management experiences were identified and so too were barriers to adaptive coping behaviour and actions or facilitators that might be usefully address these challenges. The findings from this study suggest the optimal pain management programme is one that (i) is widely

accessible, (ii) is supported or guided by credible healthcare professional, (iii) is dedicated to both the child and parent user, (iv) may be tailored to both the child and parent user over time (i.e. as support needs and treatment goals change). Findings from the current study demonstrate that the experiences of children and parents coping with paediatric chronic pain may be conceptualised within the SCT framework and correspond to the SCT constructs of self-efficacy, outcome expectations, self-regulation, observational learning, incentive motivation, behavioural goals and reinforcement. These findings are useful to inform intervention development and may guide future research.

Chapter 5 Development and usability of an Internet based intervention for paediatric chronic pain management: A mixed methods study (Study 3)

5.1 Chapter overview

This chapter describes initial usability testing of the adapted, online version of the *Feeling Better* pain management programme. This chapter will describe the collaborative development of each iteration of the web-based *Feeling Better* intervention using feedback from potential end-users and experts in pain management and intervention development. The methodological approach to this research is briefly outlined and described in greater detail in Chapter 2. This study is part of an overall body of research that has been conducted in four phases and has produced four work-packages.

5.2 Introduction

The findings from Study 1 and 2 support the assertion that Internet-mediated interventions may be one solution to current access and resource issues that prevent access to psychological support for paediatric pain management. Study 1 identified the most commonly included components in existing, effective ICT-based interventions for paediatric pain management and Study 2 highlighted psychological determinants of behaviour and associated intervention components most likely to engage this age specific cohort. There is a paucity of studies that explore the ICT-based treatment response or psychosocial experience of chronic pain in school age children. The findings from both studies highlight a lack of understanding of the design features that may be associated with more effective interventions. Thus, where Study 2 explored end-user psychological support needs and preferences, Study 3 explores the design features and intervention components most likely to engage children under 12 years of age with chronic pain. Study 3 builds on these findings by illustrating the usability of an intervention that combines evidence, theory

and end-user input.

5.2.1 Barriers to treatment

Evidence-based practice recommends the provision of support for chronic pain as soon as non-coping occurs, to prevent maladaptive behaviour becoming entrenched and the development of co-morbidities (Scottish Intercollegiate Guidelines Network [SIGN], 2013; Palermo et al., 2012). However, the realities of clinic-based practice prevent this (Children's Rights Alliance, 2010). The pain journey from diagnosis to treatment and subsequent adjustment is not typically clear cut. Children with chronic pain may be diagnosed early or they may be required to adjust to their condition before they have a name for their condition as is the case for many who experience long waiting times and undergo batteries of diagnostic tests. Cognitive behavioural therapy for chronic pain management is associated with small to moderate effect sizes for improvement in a range of clinical variables associated with chronic pain. However, many chronic pain patients do not have access to psychological support for pain management (Children's Rights Alliance, 2018). The provision of CBT is often restricted by a lack of access to qualified healthcare professionals and treatment availability being restricted to urban locations. There is a need to improve access to evidence-based treatments for paediatric chronic pain management. Internet-based cognitive behavioural therapy may be a viable solution to address access and resource barriers.

5.2.2 Novel modes of treatment delivery

The Institute of Medicine (2011) suggested a key area for future research should be the development of treatments and strategies that address barriers to active pain coping particularly for vulnerable populations (Ehde, Dillworth, & Turner, 2014). ICT-based therapy delivery may be an acceptable, accessible, and efficient means of support for several reasons. First, young people are generally more comfortable with technology and Internet-use compared to adults, therefore the delivery of therapy online

may be perceived as more acceptable and less “psychological”. Second, online interventions overcome geographic and schedule constraints, families coping with paediatric chronic pain can learn more about the nature of chronic pain and adaptive coping from the convenience of home. Third, the anonymity afforded by online intervention may overcome the stigmas associated with accessing psychological support for pain management. This is consistent with the findings from Study 3 which found that children want to fit in with their peers, to establish a sense of belonging and normality. The requirement to practice coping skills sets them apart from their healthy friends therefore, by their own report, they may avoid practice. The privacy afforded by accessing support in the comfort of home may overcome this obstacle. Fourth, the habit of practice may be easier to instil if practice is facilitated by ready, timely access to the required information and training.

5.2.3 Acceptability

Considerable drop-out rates are noted in treatment studies for paediatric pain management (Eccleston et al., 2014b; Fisher et al., 2015). This suggests there is still room for improvement and there is a need to understand the factors that lead pre-adolescent children with chronic pain and their parents to engage with and complete an online intervention. The acceptability of ICT-based pain management support according to age and developmental level is unknown. Learning from existing usability literature is impeded by a disproportionate focus on the user experiences of adolescents or adults (Long & Palermo, 2009; Stinson et al., 2010b; Riiser, Løndal, Ommundsen, Sundar, & Helseth, 2013; Nelson, Bethune, Lagotte, & Osborn, 2016). ICT-based psychological therapies are often designed to appeal to an older age range with little consideration of the features that make an engaging and developmentally appropriate programme for younger children. To develop a treatment approach that is both widely accessible and acceptable to children with chronic pain it is important to first understand how children view and experience these novel methods of treatment delivery (i.e. technology-mediated therapy) within the context of their world experience as pre-adolescent children living with a long-term condition

(Lundy, 2007).

5.2.4 Design features and engagement

A review of the literature offers limited instruction on how best to compose and utilise web-based interventions in terms of what works and for whom. This is partly due to the exploratory nature of much of the research in the field. Interest in this topic is increasing. Emerging guidelines and empirical evidence are a welcomed contribution to our current understanding of the reach, appeal and potential of Internet interventions (French et al., 2014; Morrison, 2015; Yardley et al., 2015; Yardley et al., 2016). In a critical review of the literature Morrison et al (2012) identified four interactive design features associated with more positive outcomes in e-Health interventions: social context and support, contacts with intervention i.e. supplementary correspondence, tailoring and self-management (Morrison et al., 2012). Combined, these features suggest an effective and engaging online intervention must address the isolation or anonymity associated with the remote nature of therapy delivery (e.g. social support, supplementary correspondence and tailored advice) while also facilitating independent use and coping (e.g. self-management). A follow-up review of theory-based strategies that influence the impact and usage of digital health interventions identified tailoring, social support, self-management, the information architecture and the general intervention approach as most efficacious in enhancing intervention impact and usage (Morrison, 2015). This would seem to correspond with Ritterband's (2009) model of behaviour change in Internet interventions i.e. the clinical content and use of the intervention will have a mediating effect on outcomes. The impact of self-assessment and tailoring on engagement with online interventions was examined in a mixed methods study of participants use of an online intervention. Qualitative feedback suggested self- assessment without tailored feedback is less acceptable because the advice is not personalized. This approach was also associated with greater drop out compared to those with tailored feedback (Yardley et al., 2016). O'Connor and colleagues conducted a systematic review of qualitative studies to identify the barriers and facilitators of

patient and public engagement and recruitment to digital health interventions (DHIs). Participants included patients, care-givers, healthy adults and healthcare professionals. The authors reported that personal agency and motivation (self-efficacy), personal life and values, the engagement and recruitment approach and the quality of the DHI were all factors that influence the engagement and recruitment to DHIs (O'Connor et al., 2016).

5.2.5 Internet interventions

We know from similar interventions that eight treatment sessions are the most commonly implemented format in the pain literature (Fisher et al., 2015) and that this period of delivery may be enough to achieve change in self-management behaviour (Carnes et al., 2012). Research suggests shorter (< 8 weeks) group and healthcare professional delivered interventions may be as effective or more so than longer (4-8 months, >8 months) programmes (Carnes et al., 2012). The reasons for this are unknown but may be in response to feasibility, population characteristics or research constraints. An average treatment duration of 6.4 hours has been associated with pain reduction across illness groups and some studies have found internet assisted therapy to be as effective as face to face therapy for pain management (Kashikar-Zuck, 2010). Moreover, eight to ten treatment sessions are consistent with the number of in-person sessions typically allocated to a service user attending an outpatient psychology service in Ireland (Children's Rights Alliance, 2018). It may be that this commonality will facilitate easier comparison of findings between in-person and remotely delivered CBT treatment studies.

5.2.6 Usability testing

Usability testing is essential to the success of online interventions where user autonomy can quickly translate to pre-treatment drop-out. It is important to accurately assess functionality or other issues which might affect uptake and retention. This is particularly important step for the proposed *Feeling Better* intervention which is hosted on a purpose-built

website and designed for a relatively under-researched sub-group of the pain population. Usability is evaluated differently in each field of research. The field of Human-Computer Interaction (HCI) is replete with taxonomies detailing the criteria for quality assessment of web applications and usability is just one characteristic of the quality of the software product. Given the scope of the current study, usability was examined in terms of three quality characteristics, chosen because they are an account of usability from the perspective of the real end-user, namely functionality (i.e. technical issues), acceptability (i.e. user satisfaction) and usability (i.e. ease of use and understanding) (Fernandez, Insfran, & Abrahao, 2011). This was achieved by exploring user perceptions of all three characteristics.

5.2.7 Adapting the Feeling Better manual – consideration of developmental factors

The adaptation process was primarily guided by developmental considerations. The cognitive and emotional development of the child must be considered in terms their capacity for understanding and self-regulation (Bandura, 1989b; 1991). To address cognitive and emotional understanding we must incorporate skills to identify, label and understand the causes, consequences and interaction of thoughts, emotions and behaviours. Children begin to develop the ability to understand emotional experiences at pre-school age and by the end of this period they can appreciate their own and others emotional experiences (Dunn, Brown, Slomkowski, Tesla, & Youngblade, 1991; Denham, 1998). As children mature their cognitive abilities develop and they begin to understand multiple cues (e.g. facial and situational) (Saarni, 1999; Denham, Brown, & Domitrovich, 2010). By mid-childhood, typically developing children can engage in self-regulation strategies (Saarni, 1999). Emotional skills are highly influenced by cognitive and social development (Saarni, 1999; Bandura, 1989a). As stated previously, typically developing children in the 7-12 age range are rapidly developing logical thinking, perspective taking skills and experiencing decreased egocentrism (Piaget, 1964; Piaget & Inhelder, 1969). Logical thinking during this period is believed to be largely concrete and dependent

upon observable events (Piaget, 1964). Aside from indicating the importance of observational learning, these developmentally appropriate cognitive limitations suggest the cognitive components of the treatment approach should be facilitated using concrete methods e.g. ‘I sentences’, visual aids or worked examples of complex strategies. This may be particularly important for children with long term pain. Research examining cognitive and emotion-related skills find atypical cognitive and emotion limitations, biased perception and atypical learning, concentration and memory deficits among children with chronic pain which are implicated in the maintenance of chronic pain (Simons et al., 2014). Thus, attending to these factors may lead to better treatment outcomes. Therefore, we needed to devise creative ways to engage young children in the intervention, particularly in the more challenging cognitive components of the *Feeling Better* intervention. This means incorporating stories, interactive puzzle-like exercises, computer-game terminology and graphics in the presentation of treatment components. We invited collaboration where possible e.g. in the choice of fun tasks or joint development of meaningful goals (Friedberg & McClure, 2002).

5.2.8 Aims

The aim of this study was to conduct a preliminary evaluation of the *Feeling Better* intervention prototype in terms of acceptability and usability. Specifically, this study is designed to answer **Research question 3: *How do younger children with chronic pain (5-12 years) and their care-givers perceive ICT-based therapy delivery and what are the design features and intervention components most likely to engage this population?***

5.2.9 Objectives

Specific objectives included: (i) identification of barriers and facilitators to the uptake and utilisation of a web-based, self-management programme and (ii) establishment of a link between user preferences, support needs and the content and design features of the programme.

5.3 Methods

5.3.1 Theoretical framework

This process involved an in-depth needs assessment of children with chronic pain and their care-givers, the selection and application of theory-based methods and practical strategies and iterative, person centric programme planning in terms of intervention and website characteristics (Chapters 2-4). As stated in Chapter 2 SCT guided the adaptation and intervention website development process. CBT strategies that utilised relevant mechanisms of behaviour change were selected for implementation in the *Feeling Better* prototype. This guided the selection of intervention components e.g. behaviour change techniques which in turn informed the development of design features (website and mode of delivery features). End-user input was used to validate these theoretical considerations and further inform intervention website design.

5.3.2 Copy editing

Text-based content was adapted with guidance from the National Institutes of Health checklist for the development of easy to understand print materials for low-literacy audiences (NIH, 1998). Which recommend limiting the number of concepts per treatment session, removing jargon and writing in 'active voice'. Layout recommendations include the use of simple headings and sub-headings, large font, underlining and bolding content where appropriate. Condensing a body of work such as the *Feeling Better* manual required a systematic approach to keep track of decision making and account for the inclusion or exclusion of materials and this is described further below.

5.3.3 Prioritisation process

The MoSCoW Prioritization method (Clegg & Barker, 1994) is an established, simple process of extracting what is valued, relevant and feasible from an expansive data source, within the constraints of a given project. This requires decision-making about content in terms of what the

project ‘must have’, should have, could have or won’t have time for. The MoSCoW method is specific and focused on the end- product e.g. a condensed version of the original *Feeling Better* manual.

5.3.4 Website development

Ritterband et al., (2009) identified key features thought to contribute to the effectiveness of internet interventions for behaviour change (see Figure 5.1). This model was used as a guide to the development of the intervention website. Qualitative data were retrospectively mapped to Ritterband’s categories. These are outlined as follows:

User participation: User participation is described as the ability of the internet intervention to engage or involve the user through interaction (e.g. interface with the system which produces cause and effect action), reinforcements (e.g. rewards) and testing (e.g. games, quizzes) (Ritterband et al., 2009). **Delivery:** Delivery refers to the ways in which the intervention is delivered i.e. specific modes of delivery such as animation, audio, illustrations/graphics, text, video and vignettes. **User satisfaction:** User satisfaction considers the credibility, style and likeability of the internet intervention (Ritterband et al., 2009). **Appearance:** This refers to the look and feel of the Internet intervention including colour, screen size and organisation of content. **Behavioural prescriptions:** This category refers to the content within the website which instruct the user on what to do to address the problem. **Support:** Support may be understood as external human support facilitated by the system in the form of personalised correspondence or even adjunct face to face meetings. **Assessment:** Assessment refers to the ability of the programme to measures the needs of the user and provide tailored clinical content and recommendations. **Burdens:** The burdens of using an Internet intervention are specific to the content of the intervention and not technological or environmental obstacles. For example, difficulty of use that includes intervention length or poor application navigation (Ritterband et al., 2009).

The terms tailored and personalized are used interchangeably in the literature and by participants in the current study. Here a tailored

intervention is one that introduces clinical content based on the needs of the individual or patient group based on baseline assessment information. A personalized intervention is one that refers to the individual user's name (i.e. self-referent cue) and personally relevant input (e.g. self-selected goals) but provides clinical content based on the support needs associated with the health problem in general.

In addition, the design features i.e., modes of delivery incorporated in the *Feeling Better* intervention are illustrated using Webb et al (2010) classification scheme. Optional modes of delivery such as navigation functions, entertainment value and credibility of source previously suggested by Webb and colleagues (2010) were also included in analysis of the *Feeling Better* intervention.

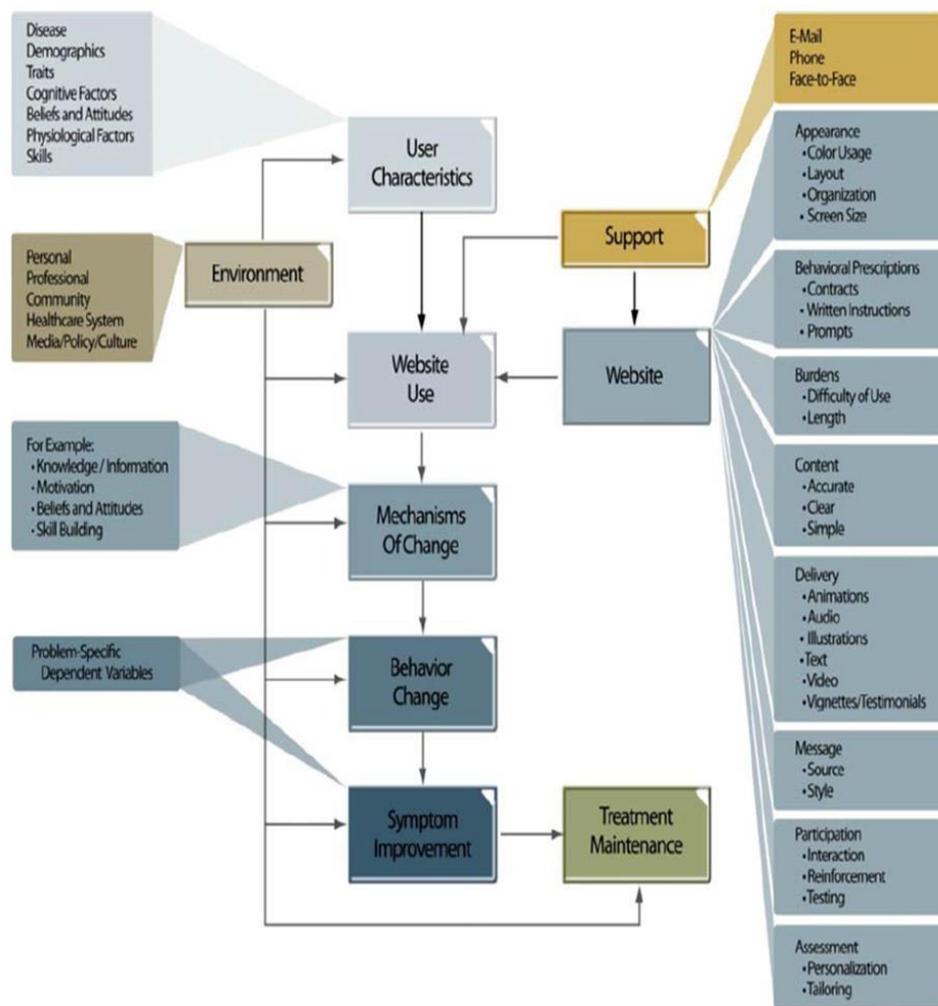


Figure 5.1 Ritterband's Behaviour change model of internet interventions

5.3.5 Participants

To develop the *Feeling Better* intervention and to test the usability and acceptability of each iteration of the *Feeling Better* website, we recruited English speaking children with chronic or recurrent pain and their parent(s) from the community. Overall, twenty-four children and thirty-four parents were involved in the design and development process. A total of eleven children with juvenile idiopathic arthritis and twenty-one parents took part in separate PRP workshops focused on the lived experience of chronic pain management. These workshops closed with a ‘think-aloud’ group evaluation of an initial draft of the *Feeling Better* intervention website (Iteration 1). A total of five children and five parents took part in the second round of PRP prototype design workshops focused on the intervention appeal, relevance and ease of understanding (Iteration 2). A further eight out of fifteen consenting parent child-dyads took part in online usability testing and completed website evaluation questionnaires (Iteration 3) (see Table 5.1).

5.3.6 Recruitment and eligibility

Using convenience sampling, children aged 5-12 years with chronic pain were invited to participate in the various phases of this research via communications sent to online, community-based parent support networks, clubs and organisations providing support and services for children located in the Republic of Ireland. Prior to data collection in each phase of development, participant information sheets, parent consent forms and children assent forms were distributed by researchers to parents who had expressed an interest in taking part (Appendix 15 and 16). Logistical issues determined the testing site for each iteration of website development. The first iteration was evaluated at two sites, namely, a Family Fun Day event organised by a parent support network (Arthritis Ireland) and at a meeting of parents and children invited to the School of Psychology building. The second iteration was tested online and evaluated in a group setting at the home of one of the participants. The third iteration was tested and evaluated online using a purpose-built website and online survey. The Research Ethics

Committee at the National University of Ireland Galway approved all study procedures prior to participant enrolment.

Eligibility criteria for the study included:

- Age (5 to 12 years) and resident in the Republic of Ireland
 - Doctor diagnosed non-malignant chronic pain or chronic condition in which pain is a primary symptom (self-reported)
 - Proficient in English
 - Reports the presence of chronic or recurrent pain for more than 3 months
 - Reports pain-related disability or psychological distress
- Exclusion criteria:
- Reports the presence of medical or psychiatric risk factors
 - Major cognitive impairment that would impact ability to participate

5.3.7 Usability Methodology

A mixed methods usability testing approach with participatory workshops and online user- testing and survey assessment was employed. This research adheres to the consolidated criteria for reporting qualitative research (COREQ) checklist (Appendix 17). This methodology began with *Iteration 1*, a series of participatory and ‘think-aloud group workshops with two aims, namely: (i) to explore the lived experience of chronic pain self-management among pre-adolescent children with chronic pain and their care-givers and (ii) to gain insight into potential user perceptions of health technology as a means of treatment delivery. To achieve these aims we used participatory methods and theoretical thematic analysis (Braun & Clarke, 2006) to identify categories of experiences that might be addressed using an online, self-management treatment approach. At the close of these workshops we conducted a ‘think-aloud’ session in which we asked the assembled children and parents to review stand-alone prototype webpages which were demonstrated on a laptop for child group 1(CG1) and projected

onto the wall of the workshop venue for child group 2 (CG2). At this stage, the researcher guided potential users through the prototype webpages and prompted responses to open-ended questions on the aesthetics, readability, content and specific modes of delivery. Children (n = 11) and parents (n=21) offered feedback on the look and theme, layout and structure and content and readability of these early drafts. Participants were encouraged to think-aloud and comments were recorded, transcribed and analysed qualitatively.

Iteration 2: The second phase of website development involved participatory workshops focused on design and usability issues. In this second phase of intervention development participants were asked to evaluate a revised prototype of the Feeling Better website. The prototype contained four treatment modules and participants were given two weeks to browse the website prior to taking part in the participatory workshop. Separate workshops (30-40 minutes each) were carried out with children with various types of juvenile idiopathic arthritis and their care-givers. During each workshop, participants were guided through a series of activities to explore (i) barriers and facilitators to uptake and utilisation of technology to support pain self-management and (ii) design ideas and preferences for the website platform to support children with chronic pain in a relevant, practical and engaging way. A participatory research process approach was employed. Phase I of the PRP workshop involved participants in the generation of data in the form of written responses or ideas on coloured pieces of card, in response to a research question defined by the researcher at the start of each workshop. Phase II of the PRP involved participants in the categorisation of their data into groups of ideas and responses based on saliency i.e. most frequent and most important. Finally, Phase III of the PRP involved participants in the creation of a schematic map of their categorised ideas in order of importance. Children and parents were asked to respond to a single question:

“If you (your child) takes part in this programme, what would it need to have to help you (he or she) to cope with pain?”.

In their separate groups, children and parents then created a ‘web of ideas’ of what they felt was suitable content and mode of delivery

components. Participants comments were recorded, transcribed and analysed qualitatively. Data collected from this process also included written responses, schematic concept maps and field notes.

Iteration 3: The third phase of development involved staggered online user-testing and evaluation of a revised, final version of the *Feeling Better* intervention website. This research was conducted with a further eight child-parent dyads who completed user testing of the website functionality. This involved user-testing of support features e.g. internal email messaging system, homework submission and completion or of interactive content features at least once. At the close of each of eight modules, participants were asked to rate the module completed. This involved a series of qualitative open-ended questions based on Ritterband's behaviour change model for internet interventions (Ritterband et al., 2009). Upon completion of the overall website, all participants were asked to complete an online survey assessment of their user experience. The online survey included demographic characteristics and the Internet Evaluation and Utility Questionnaire (Thorndike et al., 2008; Ritterband et. al., 2008). The Internet Evaluation and Utility Questionnaire is a standard validated 5-point Likert scale which measured participants' experiences and perceptions of an Internet intervention with higher score signifying more favourable responses. Earlier and shorter version of this measure revealed good internal reliability ($\alpha = .69$) (Thorndike et al., 2008; Ritterband et. al., 2008). Concurrent with this process, the intervention website was also reviewed for accuracy, relevance and developmental appropriateness by the design team which included two clinical psychologists (BMG and JE) and by an informal group of experts in intervention development and paediatric pain management. Data collection focused on the categories of data that would inform website development, these included: (i) the desired features and characteristics of a self-management support platform and integrated web-app (online pain diary) and (ii) participant identified barriers and facilitators of pain self-management using digital health technology. The quantitative and qualitative data collected informed the final iteration of the website and integrated web-app.

5.3.8 Data Analysis

The structure and functionality of the *Feeling Better* website was developed in line with the main categories of Ritterband's behaviour change model for internet interventions (Ritterband, 2009) and a mode of delivery classification scheme (Webb et al., 2010). As such these frameworks were used to guide the analysis of qualitative usability data and facilitated the translation of findings to relevant website design features which were included in the final iterations of the website. Where possible, audio-tapes from 'think-aloud' and participatory group workshops were transcribed verbatim after each iteration. Field notes were integrated with transcripts. Transcripts were analysed using simple, qualitative content analysis. Transcripts were read independently by two investigators (AT, SOH) and coded in line with intervention development framework categories. Quantitative data from participant questionnaires were analysed using SPSS 17.0.

Analysis of data produced in each iteration of the development process was also guided by the research questions driving each work-package. The findings from this study build on those of work- package 2 by linking child and parent reported experiences of pain management to evidence-based behaviour change constructs and intervention website components. To link the user experience with practical modes of delivery, the findings from work-package 3 were retrospectively mapped to the main categories of Ritterband's behaviour change model for internet interventions (Ritterband et. al., 2009). As with the previous study, the current data analytic approach is influenced by the central tenets of participative research and a phenomenological approach to understandings the user experience (Braun & Clarke, 2006).

5.4 Results

The findings from this mixed-methods study were used to inform an ongoing intervention development and evaluation process and to generate a list of desired Internet intervention components that might be applied to better engage younger children with chronic pain in a pain self-management

intervention. This is illustrated in the following results. Section I is a presentation of the findings from each iteration of intervention development, Section II is an interpretation of findings in terms of Ritterband's (2009) model of behaviour change in Internet Interventions.

5.4.1 Section I

5.4.2 Participant Characteristics

In total, 24 children and 34 parents contributed to the intervention website development process. A total of 58 contributed ideas and opinions on intervention website development in the preceding qualitative study (Study 2) and the current mixed-methods, usability study (Study 3). For the current usability study, among eligible participants who initially expressed an interest in taking part in the various phases of intervention development, approximately 15 (38%) children and 5 (13%) parents passively withdrew prior to participation. The mean age of the child participants was 9.9 years ($SD = 1.57$) and the mean age of parent participants was 42.5 ($SD = 2.81$). Tables 5.1 and 5.2 present participant characteristics, illustrating which think aloud, PRP workshop or survey group they participated in. Participants were recruited through several social media groups: Arthritis Ireland (AI); the Irish Children's Arthritis Network (iCAN); the ehlers-danlos syndrome (EDS) parent support network.

Iteration 1: Participatory and 'Think-aloud' Workshops (Study 2 and 3): The average age of child participants ($n= 11$) was 9.9 years ($SD= 1.64$; range 7-12) (see Table 5.1). The average age of parent participants ($n=21$) was 42.9 years ($SD = 2.58$, range 38 to 47) (see Table 5.2).

Iteration 2: Online user-testing and Participative Research Process Design Workshop (Study 2 and 3): The average age of child participants ($n=5$) was 10 years ($SD = 1.58$; range 8-12). All five child participants were diagnosed with a type of JIA (self-reported). One child participant was female and 4 were male (see Table 5.1). The average age of parent participants ($n = 5$) was 41 years ($SD = 4.09$; range 35 to 46). (see Table

5.2). All five parent participants were female. Participants were recruited from the social media forums associated with the iCAN (n=2) and AI (n=3).

Iteration 3: Online User-testing (Study 3): The average age of child participants (n=8) was 10 years (SD = 1.69, range 8-12) (see Table 5.1). Two child participants were diagnosed with JIA and three child participants were diagnosed with EDS. Two child participants were female and 3 were male. The average age of parent participants (n=8) was 42.5 years (SD = 2.67, range 39 to 47) (see Table 5.2). All five parent participants were female.

Twelve children and parents were approached by the research team and agreed to participate in user-testing of the online prototype. A total of 8 child-parent dyads (67%) took part and fully completed online user-testing. Some parents volunteered reasons for non-participation. Reasons included: “My daughter is doing well now; I don’t want to rock the boat”, “We have just finished a course with Arthritis Ireland” and “I’m already taking part in another study”. Participant characteristics are presented in Tables 5.1 and Table 5.2. Three parent-child dyads agreed to take part but dropped out. The only reason for drop-out offered by one parent was “lack of time”. As drop out occurred prior to completion of the first module, data from these participants were not included in analysis.

Iteration 4: Expert Feedback (Study 3): Three experts in paediatric pain, intervention development and pain management were approached for feedback on the prototype Feeling Better programme. Their feedback was used to refine the prototype prior to testing in the feasibility trial.

Table 5.1

Demographics of child participants in think aloud (TA1&2), participative (PRPG3) and survey groups (SG4)

Participant number	Sex	Age (range)	Child Pain Type	Pain Duration (years)	Child-TA1	Child-TA2	Child-PRPG3	Child-SG4
1	F	11	E-oligo	4	*			
2	M	7	P-oligo	1	*			
3	F	9	P-oligo	1	*			
4	F	8	Poly Rf-	1	*			
5	F	9	Poly Rf-	2	*			
6	F	12	Poly Rf-	3		*		
7	F	12	Enthesitis- related	5		*		
8	F	11	E-oligo	3		*		
9	F	11	Enthesitis- related	5		*		
10	F	10	P-oligo	4		*		
11	F	9	Poly Rf-	3		*		
12	M	12	Poly Rf-	4			*	
13	M	9	P-oligo	2			*	
14	M	10	Poly Rf-	2			*	
15	F	8	Poly Rf-	1			*	
16	M	11	E-oligo	3			*	
17	F	8	P-oligo	2				*
18	F	12	E-oligo	3				*

19	F	11	Poly Rf-	3	*
20	F	9	Poly Rf+	2	*
21	F	12	Systemic	2	*
22	F	8	EDS	1	*
23	F	9	EDS	2	*
24	F	11	EDS	1	*

EDS: Ehlers Danlos Syndrome, E-oligo: Extended-oligo- articular juvenile idiopathic arthritis; P-oligo: Poly-articular juvenile idiopathic arthritis; Poly Rf: RF-positive poly-articular juvenile idiopathic arthritis. Enthesitis-related: Enthesitis-related juvenile idiopathic arthritis; TA1: Think aloud Group 1; TA2: Think Aloud Group 2; PRPG3: Participative Research Process Group 3; SG4: Survey Group 4

Table 5.2

Demographics of parent participants in think aloud (TA1&2), participative (PRPG3) and survey groups (SG4)

Participant number	Sex	Age (range)	Parent-TA1	Parent-TA2	Parent- PRPG3	Parent-SG4
25	F	43	*			
26	F	45	*			
27	F	45	*			
28	F	46	*			
29	F	42	*			
30	F	47	*			
31	F	40	*			
32	F	42	*			
33	F	41	*			
34	F	45	*			
35	F	40	*			
36	F	45	*			
37	F	44	*			
38	F	43	*			
39	F	39	*			
40	F	41	*			
41	F	45		*		
42	F	46		*		
43	F	40		*		

44	F	38	*		
45	F	43	*		
46	F	35		*	
47	F	46		*	
48	F	42		*	
49	F	43		*	
50	F	40		*	
51	F	40			*
52	F	42			*
53	F	39			*
54	F	44			*
55	F	45			*
56	F	41			*
57	F	42			*
58	F	47			*

EDS: Ehlers Danlos Syndrome, E-oligo: Extended-oligo- articular juvenile idiopathic arthritis; P-oligo: Poly-articular juvenile idiopathic arthritis; Poly Rf: RF-positive poly-articular juvenile idiopathic arthritis. Enthesitis-related: Enthesitis-related juvenile idiopathic arthritis; TA1: Think aloud Group 1; TA2: Think aloud Group 2; PRPG3: Participative Research Process Group 3; SG4: survey Group 4

5.4.3 Iteration 1 Think aloud walk-through - group comments by type

In Iteration 1, the same questions were asked of children and parents: *“What do you think of pain management support delivered using the Internet? / “What do you think of this method of delivery (Feeling Better programme examples)”*. Children (Child TA1) and parents (Parent TA1) suggested a “individualised” programme, tailored to individual or group needs (e.g. JIA-associated chronic pain); using a “personal profile” with inspiration pictures: “visuals of comfort”; a support programme that is “interactive”, shows “progress”, facilitates a social network of support; e.g. “being able to connect online with other children” and provides access to experts e.g. “Q&A with experts”; includes parents – “giving them the information they need to work as the back-up” and teaches in a way that appeals to the child e.g. “make it about us, not just arthritis” (Child TA1; girl, age 11).

5.4.4 Iteration 2: PRP user-testing - group comments by type

In Iteration 2, the same questions were asked of children and parents: *“If you (your child) were to use this website / take part in this programme - what would it need to have to keep your interest / help you (your child) to cope with pain?”*. Figure 5.2 and 5.3 represent a quantitative summary of combined user comments generated in the PRP group workshops. Responses were categorised by their content, namely positive comments, negative comments or suggestions for improvement. A total 67 responses were generated by children and 37 responses generated by parents. A total of 21 (30%) responses generated by children and 28 (76%) of the responses generated by parents were positive. A total of 6 (9%) responses generated by children and none of the responses generated by parents were negative. 41 (61%) responses generated by children and 9 (24%) responses generated by parents contained a suggestion for improvement. This pattern of responses suggests child participants felt comfortable critiquing the website in front of the researcher however parents may not have felt as comfortable critiquing the website in front of the researcher.

In terms of the categorisation of data by participants, the following

agreement was reached. Comments referring to website design features would be distinguished from comments referring to visual appeal based on the function. For example, if a comment refers to a feature of the website that serves a therapeutic purpose or function e.g. goal setting it was categorised as a website design feature (i.e. referring to design or content). If a suggestion for improvement e.g. include more pictures was made with no reference to function other than assumed aesthetic appeal, it was categorised as visual appeal.

Most of the comments made by children referred to the visual appeal of the programme which was discussed in terms of how it might be improved to engage more children with the specified age group (5-12 years). The second largest category was the positive comments relating to the website design features and content. The third largest category was the suggestions for improvement in terms of the design features and content that might engage the target population. Among parents, the largest category was the positive comments referring to the incorporated design features and content. The second largest category was the positive comments regarding the structure of the programme. Suggestions for improvement offered by parents referred to design features and content and the visual appeal of the programme.

Many of the most salient comments were categorised as suggestions to improve ease of understanding e.g. “text in different colours”, “larger writing”, “put in a list” (CG3) or as suggestions to improve user engagement and website use: “different genders”, “avatars”, “let us personalize it”, “choose your own treasure (rewards)”. Some younger children (aged 6-8 years) acknowledged the need for a parent to act as coach to complete the programme: “ok if I have Mum with me” (CG3). Parent suggestions align with those of the children, both groups recommended the inclusion of a social support facility in addition to the online tutor and the integration of the programme with standard treatment and social systems: “get school/hospital on board” and “include friends”.

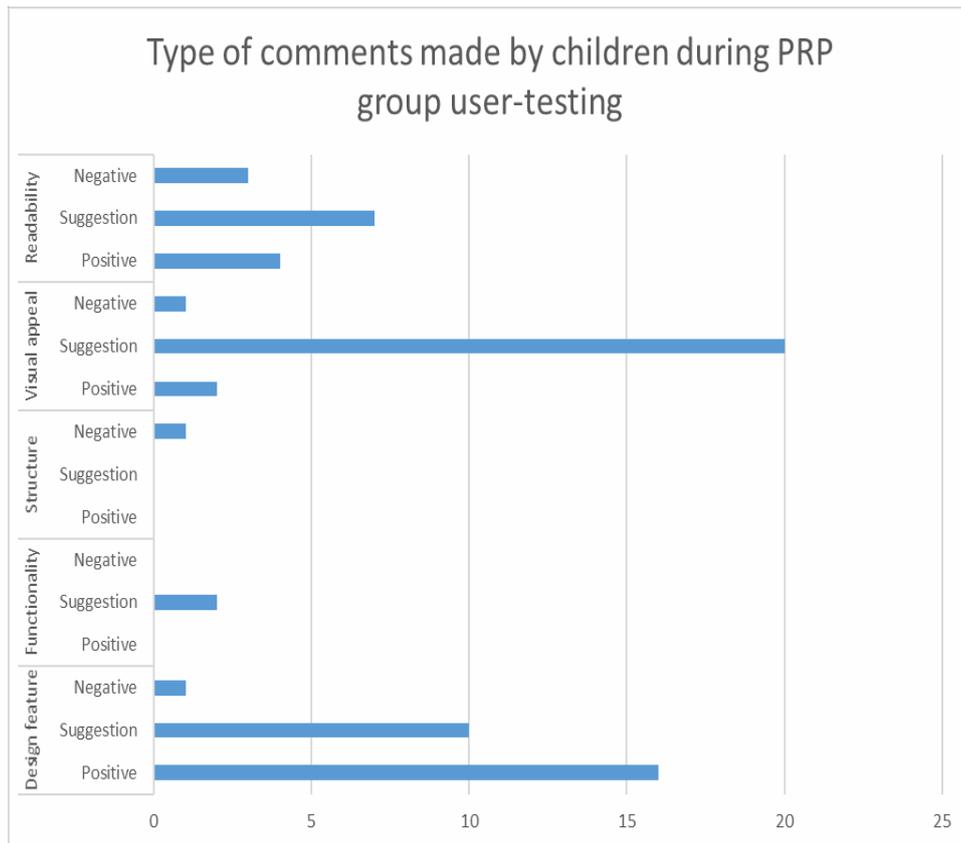


Figure 5.2 Types of comments made by children during Iteration 2: PRP Group User-testing

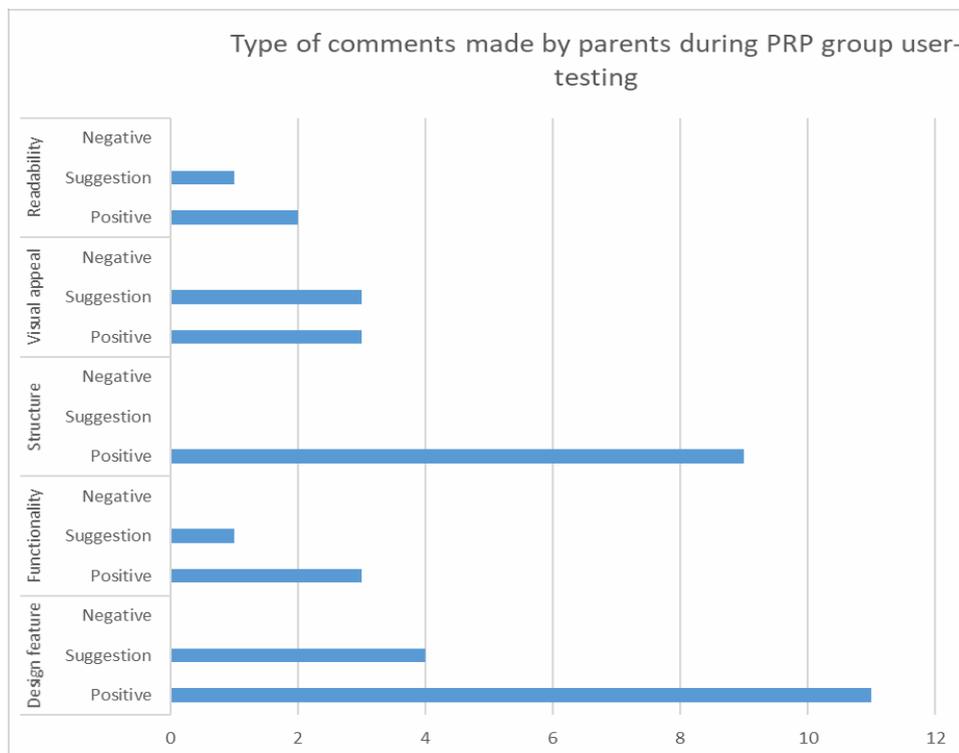


Figure 5.3 Types of comments made by parents during Iteration 2: PRP Group User-testing

5.4.5 Desired characteristics

Participant responses collected from each iteration of website development was evaluated according to the main categories of influence set out by the Behaviour Change Model for Internet Intervention (Ritterband et al., 2009) (see Table 5.3). A list of preferred design features and intervention components emerged from the overall collection of responses and design ideas offered by participants in each iteration of website development. Twelve key design features were identified are most likely to engage children aged 5-12 years with chronic pain in an Internet intervention to support pain management. Characteristics agreed by children and parents included: social support (family and peers); tailoring and personalization according to the user or group (terms used interchangeably by participants); supported use of the intervention (i.e. supplementary contact with an online tutor via email or telephone); personalized goal and reward setting, progress monitoring and feedback; novel and entertaining presentation of clinical content (i.e. visual appeal of the programme); ease of use in terms of function and access (i.e. usable and accessible via portable devices) (see Figures 5.4 - 5.6).

Children prioritised the visual appeal of the programme, tailoring and personalization; goal and reward setting, progress monitoring and feedback; updated information as the intervention progresses (i.e. feed-forward system of treatment delivery with each treatment session building on the input in the previous); tunnelled navigation and “resume later” usability options (see Figures 5.4-5.5). Parents prioritized: clinical content specific to “pain and mood”; integration with family systems (i.e. involvement or parents); integration with social systems (i.e. collaboration with hospital or school support teams); “get school/hospital on-board”, the promotion of pain self-management and reduction of dependent coping behaviour: “independent”, social support and personalization and ease of understanding (see Figure 5.6)



Figure 5.4 Iteration 3: Child PRP Group, Schematic map illustrating desired components (n=5)

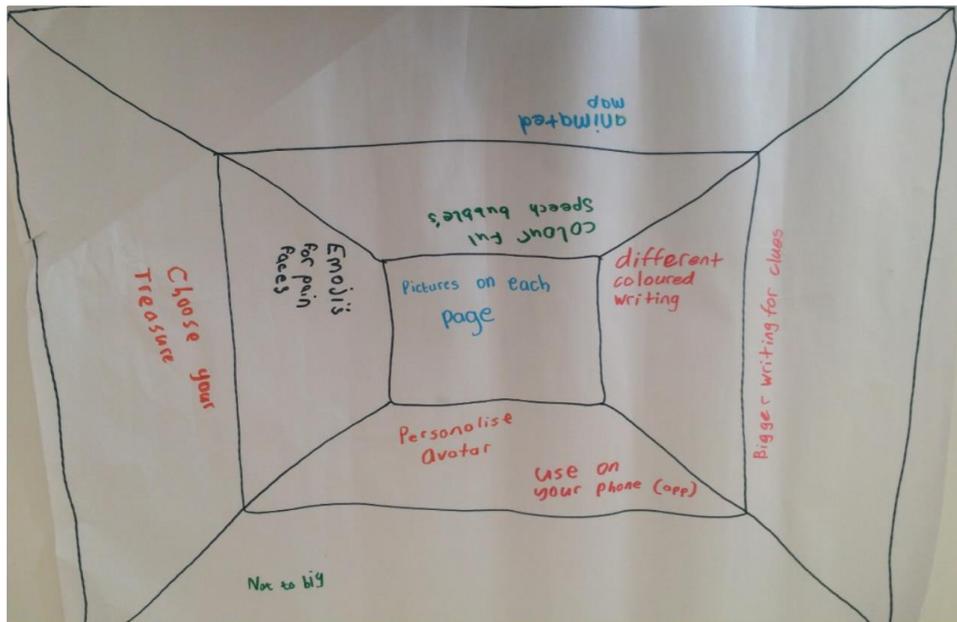


Figure 5.5 Iteration 3: Child PRP Group, Schematic map illustrating desired components (n=5)

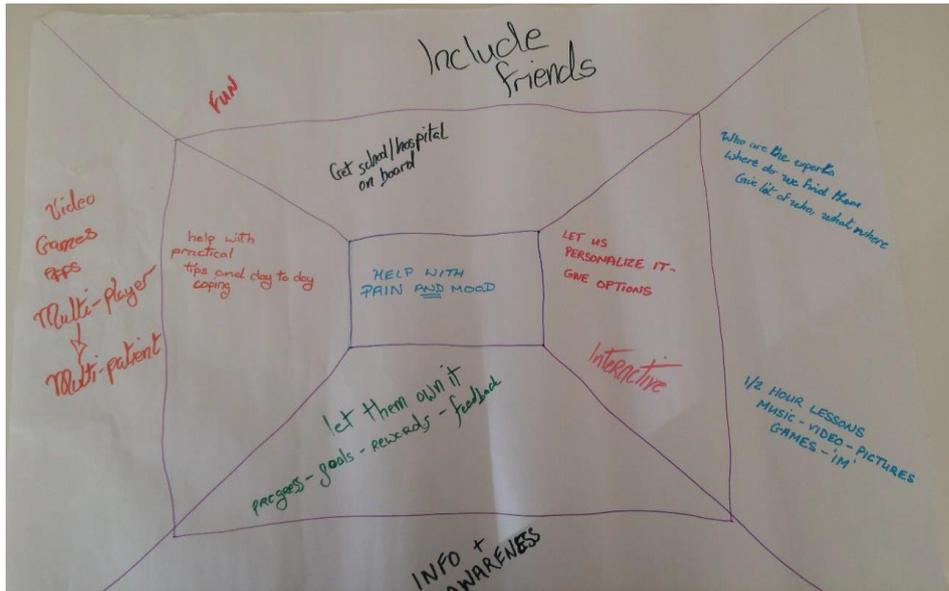


Figure 5.6 Parent Group 3, Schematic map illustrating desired components (n=5)

Table 5.3

Summary of participant comments by theme

Theme	Comments by participants	Design feature
Support: personalised correspondence could bolster confidence and engagement in the programme	“(have) someone giving you encouragement” (CG1), “peer to peer support: she’ll try anything if it means she can be like her friends”; “Q&A with experts”; (coach) “give reassurance”; “have access to an expert and other children” (PG2);	Tailoring/Personalisation/Social support. Individual user-names and pre-defined goals presented throughout the programme. Online tutor support encourages use of support services and people.
Appearance (the theme, layout and visual appeal of the website):	“more fun facts” (CG3), “different colours (text)” (CG3), “like to upgrade character” (CG3), (characters) “same genders” (CG3), “dress avatar” (CG3), “upgrade avatar after each level”(CG3), “use tick for quiz answers” (CG3), “good talking with coach” (CG3), “more pop-ups please” (CG3), “more drop-downs” (pre-defined lists of options) (CG4), “keep the fun facts” (CG3)	Novel/Game-like graphics Appealing theme, colour and interactive game-like graphics and terminology are presented throughout to enhance user experience and participation.
Behavioural prescriptions (level of comfort with language, concepts, readability):	“(programme) for kids 6-8” (CG3), “pirate post (internal message centre) better for older children” (CG3),“(change) different name for 'I sentences'” (CG3), “more labels please” (instructions for use) (CG3), “(ease of use) ok if Mom with them” (CG3), “(ease of use) have mum with them”, “bigger writing for clues” (CG3),	Graphics / Illustrations. Stories, analogy and diagrams are used to clarify the function of CBT strategies
Burdens: specific to content, not technological or environmental	" too much writing" (CG3); “is better only doing one lesson at a time” (PG3), “liked that it’s so easy to follow” (PG3), “completing one mission at a time worked for mine” (PG3),	Lower reading level. Text based instructions are simplified
Content: treatment information, clinical content	"include parents – give them the information they need to work as back-up"; "parents can offer positive comments" (PG2), “liked that prog helps them understand why”	Guided self-management Focus on enhanced function rather than symptom

	(PG3); “think it covers a good amount of topics (PG3), “very informative, quiz was good fun” (PG3), "I liked picking thoughts and feelings"; "I liked setting goals", (liked) "spotting things that make pain better or worse" (CG4),	reduction; promote benefits of active coping;
Delivery: ways in which content can be delivered (modes)	“liked cartoons” (understanding pain) (CG3), “liked the relaxation videos” ((PG3), "keep the quizzes"; "keep the map of body" (CG3);	Novel, entertaining delivery Strategies can be accessed through interactive features, pdf instruction, video and audio materials
Message: source and style of content	(provide more reassurance of data protection) "security" (PG1), “fun and informative” (PG3),	Credibility. Emphasise affiliation with NUI Galway and expertise of team
Participation: programme's ability to engage (interaction), cause and effect actions	"I liked joining crew" (promise of commitment); “I liked being able to do it myself” (CG4); “like that I can check the system without drawing her attention to her pain” (PG3), “My child would like their own password, make them feel more independent” (PG3),	Independent use Participants given autonomy to engage with the programme at their convenience, also receive reminders to use
Reinforcement: feedback / reward-based components	I liked moving up the ranks (CG4), "I liked getting more treasure" (CG4), “show progress - that'll keep her on it"(PG1), “Let parents pick the rewards” (PG3),	Incentivisation. Incentive usage through promotion of avatar based on pirate theme.
Assessment: ability to personalise /provide tailored content	“make it about us, not just arthritis” (CG1), “have a personal profile” (CG1), “individualise it” (PG1),	Personalisation. Personalisation is a key motivator

5.4.6 Iteration 3: Survey group usage Remote user testing

Parent (Parent SG4) and child (Child SG4) user ratings of website use, ease of understanding, usefulness and relevance, and aesthetic appeal were moderate to high for each of eight modules (see Table 5.4-5.6). There was some variation across modules with the relaxation training, thoughts and feelings, problem solving and relapse prevention receiving the lowest ratings for ease of understanding. This is consistent with participant responses to open-ended questions asking for the qualitative feedback on the programme (see Table 5.7). Usage data suggests the average number of days' users logged onto the prototype website was 1-2 days each in the two-week testing period (Iteration 3). A total of eight children and eight parents took part in user-testing of each of the eight original modules which comprised the penultimate iteration of the *Feeling Better* prototype. As development was ongoing, this phase of user testing was stepped meaning that user-testing was undertaken in stages as each module was released. This meant the remote-testing period spanned approximately 7-8 months. Parents and children were asked to make use of all interactive features. In total, four email messages were sent by parents and two email messages were sent by children using the internal messaging system *Pirate Post*. All messages were friendly and introductory in nature. Some parent participants enquired about how to use certain features e.g. the thought challenging exercise, the number of open, interactive fields they should complete or how to move onto the next module. One homework assignment was submitted by three participants.

Table 5.4

Children and Parent (n=8 dyads) Ratings of specific Feeling Better Modules

	How easy to use?	How easy to understand?	How useful / relevant?	How aesthetically appealing?
	0–5 (not at all–very)	0–5 (not at all–very)	0–5 (not at all–very)	0–5 (not at all–very)
	M (SD)	M (SD)	M (SD)	M (SD)
Children's Modules				
Mission 1: Understanding Pain	3.50 (0.54)	3.00 (1.07)	3.88 (0.35)	3.50 (0.76)
Mission 2: Relaxation Training	3.63 (0.52)	2.75 (0.89)	4.00 (0.00)	4.00 (0.00)
Mission 3: Exercise & Activity Pacing	3.38 (0.74)	3.25 (0.46)	3.88 (0.35)	3.88 (0.35)
Mission 4: Attention Management	3.88 (0.35)	3.38 (0.52)	3.38 (0.52)	3.63 (0.52)
Mission 5: Thoughts & Feelings	3.88 (0.35)	2.75 (0.89)	3.38 (0.52)	3.75 (0.46)
Mission 6: Problem Solving	3.13 (0.35)	2.88 (0.35)	3.13 (0.64)	4.00 (0.00)
Mission 7: Communication Skills	3.50 (0.54)	3.13 (0.35)	3.00 (0.00)	3.25 (0.46)
Mission 8: Relapse Prevention	1.63 (1.41)	2.63 (0.52)	2.63 (0.52)	2.38 (0.52)

Table 5.4

Children and Parent (n=8 dyads) Ratings of specific Feeling Better Modules continued....

	How easy to use? 0–5 (not at all–very)	How easy to understand? 0–5 (not at all–very)	How useful / relevant? 0–5 (not at all–very)	How aesthetically appealing? 0–5 (not at all–very)
	M (SD)	M (SD)	M (SD)	M (SD)
Parent's Modules				
Mission 1: Understanding Pain	3.75 (0.46)	3.38 (0.74)	3.75 (0.46)	3.88 (0.35)
Mission 2: Relaxation Training	3.63 (0.52)	3.63 (0.52)	4.00 (0.00)	4.00 (0.00)
Mission 3: Exercise & Activity Pacing	4.00 (0.00)	3.63 (0.52)	4.00 (0.00)	3.50 (0.54)
Mission 4: Attention Management	3.88 (0.35)	4.00 (0.00)	3.88 (0.35)	3.63 (0.52)
Mission 5: Thoughts & Feelings	4.00 (0.00)	4.00 (0.00)	4.00 (0.00)	3.75 (0.46)
Mission 6: Problem Solving	3.00 (0.00)	3.63 (0.52)	4.00 (0.00)	4.00 (0.00)
Mission 7: Communication Skills	4.00 (0.00)	3.50 (0.54)	3.88 (0.35)	4.00 (0.00)
Mission 8: Relapse Prevention	3.75 (0.46)	4.00 (0.00)	3.25 (4.63)	3.75 (0.46)

Table 5.5
 Children's (CG4) ratings of overall website usability: N=8

	Not at all	Slightly	Somewhat	Mostly	Very
	N (%)	N (%)	N (%)	N (%)	N (%)
How easy was the <i>Feeling Better</i> programme to use?		1(12.5)	2 (25)	5(62.5)	
How (convenient) good was it to use the <i>Feeling Better</i> programme from home?		4(50)	4(50)		
How much did the <i>Feeling Better</i> programme keep your interest and attention?		2 (25)	4(50)	1(12.5)	1 (12.5)
How much did you like the <i>Feeling Better</i> programme?				6 (75)	2 (25)
How much did you like the way the <i>Feeling Better</i> programme looked?				4(50)	4(50)
How worried were you about your privacy in using <i>Feeling Better</i> programme?	1(12.5)	4(50)	1(12.5)	1 (12.5)	
How(satisfied) happy were you with the <i>Feeling Better</i> programme?			2 (25)	5(62.5)	1(12.5)
How good of a fit was the <i>Feeling Better</i> programme for you?			1 (12.5)	6 (75)	1(12.5)
How useful did you find the information in the <i>Feeling Better</i> programme?			2 (25)	6 (75)	
How easy was the information to understand?			5(62.5)	1 (12.5)	1 (12.5)
How much did you feel you could trust the information?			1 (12.5)	6 (75)	1 (12.5)
If your pain continues or returns, how likely would you be to come back to the <i>Feeling Better</i> programme?		1(12.5)	2 (25)	4 (50)	1(12.5)
How good of a method was the Internet for delivering this programme?			1 (12.5)	6 (75)	1 (12.5)

Table 5.6

Parents (PG4) ratings of overall website usability and evaluation data: N = 8

	Not at all	Slightly	Somewhat	Mostly	Very
	N (%)	N (%)	N (%)	N (%)	N (%)
How easy was the Feeling Better programme to use?			1 (12.5)	5(62.5)	1 (12.5)
How convenient was Feeling Better programme to use?				7 (87.5)	1 (12.5)
How much did the Feeling Better programme keep your interest and attention?		1 (12.5)	5(62.5)	1 (12.5)	1 (12.5)
How much did you like the Feeling Better programme?				6 (75)	2 (25%)
How much did you like the way the Feeling Better programme looked?				7 (87.5)	1 (12.5)
How worried were you about your privacy in using Feeling Better programme?	1 (12.5)	4(50)	2 (25)	1 (12.5)	
How satisfied were you with the Feeling Better programme?			1 (12.5)	6 (75)	1 (12.5)
How good of a fit was the Feeling Better programme for you?			2 (25)	5(62.5)	1 (12.5)
How useful did you find the information in the Feeling Better programme?			2 (25)	6 (75)	
How easy was the information to understand?			1 (12.5)	6 (75)	1 (12.5)
How much did you feel you could trust the information?			2 (25)	6 (75)	
If your pain continues or returns, how likely would you be to come back to the Feeling Better programme?		1 (12.5)	1 (12.5)	4 (50)	2 (25)
How good of a method was the Internet for delivering this programme?				6 (75)	2 (25)

5.4.7 Section II

5.4.8 Modifications based on user preferences

Three/four iterative phases of website development were necessary to address challenges identified in user testing. Due to logistical limitations, i.e. access and recruitment issues, the methods employed in each iteration of website development differed from the previous. Iteration 1: established that remote modes of treatment delivery would be welcomed by younger children with chronic pain and their parents. This initial walk-through set the standard for the online approach of the programme. Iteration 2: usefully informed the intervention content and design as expected. This assessment of the prototype identified several issues relating to functionality and readability. Iteration 3: helped to refine the FB programme, highlighting barriers to uptake in a remote setting. Iteration 4: was useful to refine usability and ease of understanding further before testing in a pilot trial. Modification were carried out following each cycle of testing, a summary of which is presented in Table 5.7. Participant feedback (schematic maps and written responses on paper and online) during each iteration of website development was analysed using an open coding approach. Modifications based on user feedback and developmental considerations are presented below:

Table 5.7

Summary of problems cited by participants and subsequent modifications

	Problems and preferences cited by participants	Modifications
Iteration 1: Think-aloud group response to a walk through of the intervention using static web-pages, power-point and hardcopy storyboards	Wording too sophisticated e.g. 'frequency' "Personalise it" e.g. profile	Change words like "frequency", positive to good or better; Personalised text, greetings, messages of support
	Children and parents opted for more pictures and diagrams to convey meaning in text	More pictures, pirate themed characters and diagrams added to each module e.g. cycle of pain diagram, cartoon characters representing doctor in Communication module
	Regular access to coach	Weekly report to parents and messages of support to children
	"Involve parents"	Add instructions to involve parents, incorporate strategies that require family involvement e.g. communication skills,
Iteration 2: PRP Design Group response to a walk-through of the prototype website and prior access to the prototype (2 weeks prior to workshop)	"A lot to remember, need help following instructions"	Incorporated more automatic messages prompt and reminders
	"Change the name (of the modules) to something friendlier, less like homework"	Module names changed to Mission 1, Mission 2 Relax More - Feel Better etc.
	Children agreed they would like to see their progress through each module	Incorporated a progress bar and automatically updating progress report in each weekly check-in
	Navigation issues moving from module to module	Navigational errors corrected
	Parents suggested instant (automatic) encouragement for tasks completed	Incorporated positive reinforcement through automated messages of encouragement associated with several CBT techniques

	Simplify and explain using graphics	More pictures, pirate themed characters and diagrams added to each module
Iteration 3: Online testing/Survey Group response to remote user-testing of the penultimate prototype website	Children report preferences for more and less information	Provide information in a graded way, where user can determine depth of information required. Use images, videos where possible
	Children were confused about instructions for SMART goals	Simplified text
	3 parent participants found returning later to a complete homework quite difficult.	Created the Compass to allow participants to complete homework assignments later without having to navigate through the associated module
	3 participants found the videos difficult to stream due to poor Internet service in their area	Optimise delivery format for ease of use, include pdf -based instructions for use as an alternative
	Programme is very large	Condensed content further, larger text, fewer pages, more graphics and illustrations
Iteration 4: Expert feedback*	Make the parent pathway more interactive, engage parents in skills training	Budget constraints prevented further modifications to the parent pathway to make it more interactive.
	Replace the word homework with something more fun, less like a chore	Homework replaced with: final challenge
	Videos may be too sophisticated	Budget constraints prevented further modifications to the videos
	Communications and problem-solving module could be blended	Modules condensed but not combined in this iteration, this may be attempted at a later stage

Introduction and pain education module are too long	Introduction and pain education module were separated into Introduction and Module 1/Mission 1. Introduction module made freely accessible. Pain education module only accessible if user has consented and selected option to commit to training.
“Simplify the text”	Text simplified and spread over more pages or replaced with diagrams and graphics

*Prof. Tonya Palermo; Dr. Blake Dear and Dr. Jennifer Stinson

5.5 Interpretation of findings using the Behaviour Change Model for Internet Interventions

5.5.1 User participation (interactivity)

Interaction with the intervention website was facilitated using multiple modes of delivery. These modes of delivery varied greatly in terms of their interactivity. This included: viewing a video, listening to audio, using a “drag and drop” feature, ticking boxes, open-text writing options, drawing a link between concepts, highlighting and other forms of interaction. Navigational errors were identified by participants in Iteration 2 and corrected prior to testing in Iteration 3 (Table 5.6). Some participants reported being unclear about how to return to the home page and how they should move through the programme (PG3). In response, the “Home page” icon was emphasised, and the Instructions for Use i.e. tunnelled navigation were simplified. Collaboration and choice were offered where possible. For example, relaxation skills were made more appealing by offering the choice of location for guided visualization i.e. the beach or the forest and the option to create a personal relaxation space.

Also, the lessons that might be learned from patient case examples might be better internalized by allowing for personal reflection and the development of implementation intentions in a variety of ways. For example, following each patient narrative the opportunity to self-reflect was

introduced using “Your Story”, a strategy designed to encourage children to apply what they have just learned to their own circumstances. This typically involved asking a question about the patient case example e.g. “what does Peter do to relax?”, followed by a question that requires the child to apply the lesson illustrated in the case example to his or her own circumstance e.g. “what could you do to relax?”

The target age range is relatively large (5-12 years). One strategy to engage children from both ends of this spectrum is the facility to create goals and rewards that take individual preferences into account. This information was then fed-forward through the programme in weekly check-in, progress reviews. A pre-defined goal and reward list was added in addition to the facility to spontaneously create personal goals and rewards. This was intended to help younger children understand the task. Goals and rewards that are meaningful to the child may enhance motivation to comply with treatment. Parents were guided in how to facilitate this strategy including an example list of tangible and reasonable rewards that may be used as incentive motivation e.g. the provision of small prizes, computer games or magazines. Social rewards were encouraged e.g. favourite activity or quality time (i.e. one on one) with a parent.

Incentive motivation in the form of virtual promotions based on session completion i.e. children were promoted up the ranks from powder monkey to ship’s captain with every session completed was a favoured aspect of the intervention. In addition, Survey response data (Iteration 3) revealed that for children “getting to be captain and getting treasure”; “stories and quizzes”, “goals and rewards” and “nice messages” were the parts of the programme that were most helpful and engaging (see Figures 5.7 and 5.8).

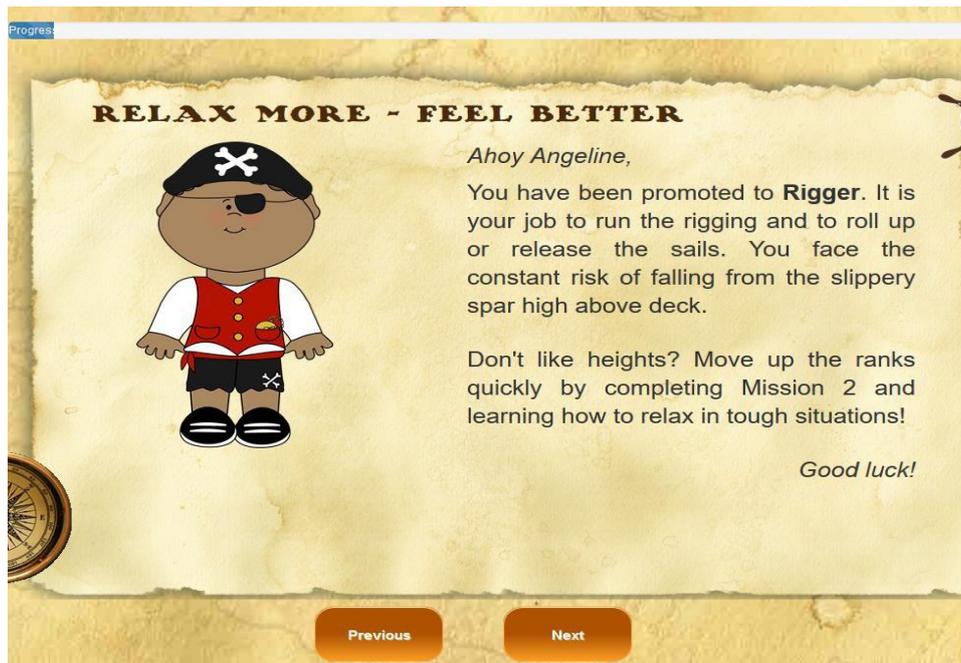


Figure 5.7 Example of virtual incentive i.e. promotion through the ranks

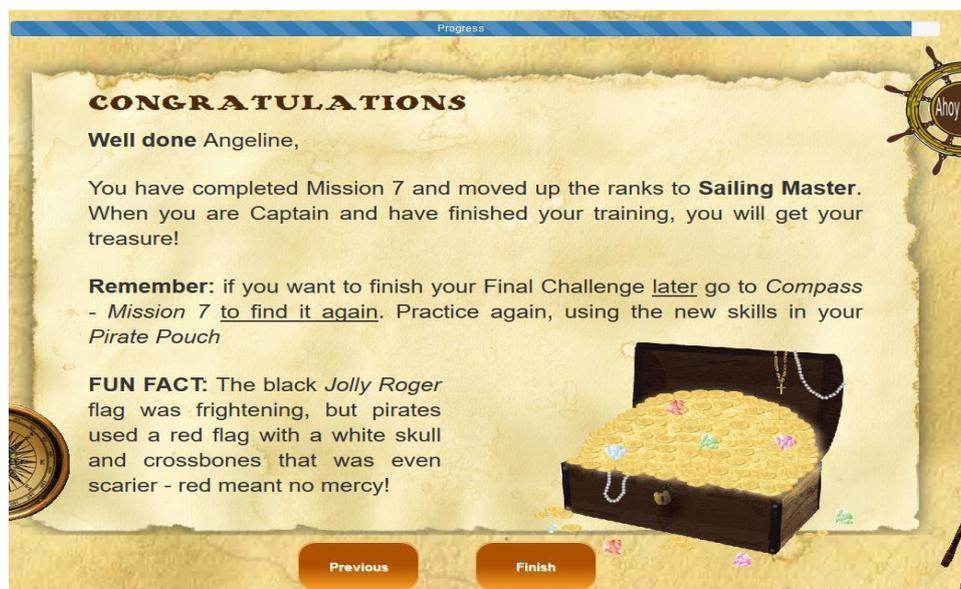


Figure 5.8 Example of virtual reward i.e. virtual promotion and accumulating treasure

Positive reinforcements in the form of automated and personalised messages of support based on progress and task completion were used to engage children and parents. Automated feedback included personalised messages of encouragement e.g. 'well done Katie you did a great' or feedback on the completion of a task delivered via the internal *Pirate Post*

messaging system or via external email (parent report) Each of these features appealed to parents and children: “like that we can see how we’re doing” (PG4; see Figure 5.9) and children suggested progress review should be included in the final intervention: “include progress reports” (n=8; CG3)

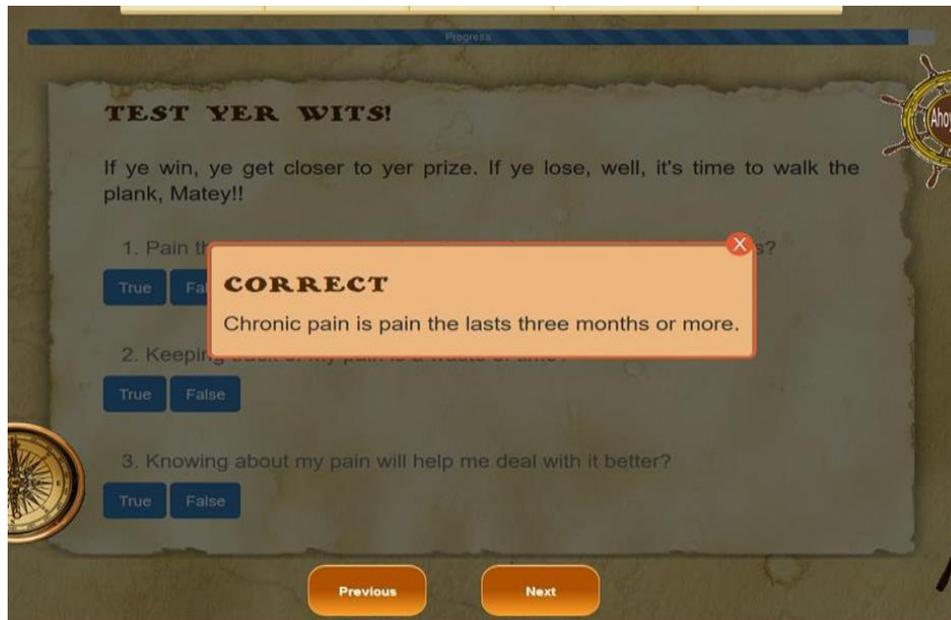


Figure 5.9 Example of automated message

Learning and behaviour acquisition was periodically tested using interactive ‘drag and drop’ games and pain education-based quizzes (see Figure 5.10). this was one of the most popular features of the programme (Iteration 2, 3 and 4).



Figure 5.10 Example of the end of module, interactive quiz

5.5.2 User satisfaction (Message likability)

When asked “how much did you like the *Feeling Better* programme?” all participants answered mostly or very (see Table 5.4 and 5.5). Most of the participant responses referring to the style and likeability of the intervention message and content were positive. This aspect of development was directly influenced by relevant literature and end-user involvement in each stage of the development process.

To accommodate emotional and cognitive factors, an effort was made to ensure the treatment components that address cognitions (i.e. beliefs or appraisals) and psychological distress (i.e. depression or anxiety) were presented in fun, enjoyable ways. For example, the *Feeling Better* programme teaches children to recognize the effects of different emotions and somatic cues on the body using interactive body-maps and “matching” exercises linking different emotions, thinking or behaviour to different body areas related to somatic cues (i.e. drag and drop or draw a line between the matching thoughts, feelings, body parts or behaviours) (Friedberg & McClure, 2002). Child report revealed children both learned from and enjoyed this technique (Child SG4).

Finally, to boost the credibility of the *Feeling Better* programme the website prominently features information linking the intervention to an academic source i.e. the National University of Ireland Galway and Centre for Pain Research Galway. This information was emphasised following feedback in Iteration 1.

5.5.3 Appearance (theme/layout/aesthetics)

The Main Page of the programme (prior to log-in) features information about the research project, contact information for the research team, a pathway to the participant information sheets and baseline assessment survey. Once a password has been issued (post-randomisation), the user is brought to the Home Page which features an information module and nine treatment sessions, an option to pick up where the user left off and a summary of progress feature which reminds the user how far they have come i.e. modules completed. A simple menu at the top of the web-page

features additional options including a Meet the Team page, a homework area which facilitates a return to module-specific homework i.e. final challenges without having to navigate to the end of each module. Children are also asked again if they are willing to take part and make a commitment to the programme. The appearance of the website was carefully designed to prevent distraction and maintain focus on the key topics introduced. The colours, layout and structure of each webpage were deliberately consistent to reduce the cognitive demands placed on participants. In terms of layout, parent and child participants opted for one key concept and five or less sentences per page. Content would then be continued to the next webpage. This was preferred even if it meant the programme would increase in size i.e. number of pages per treatment session. While personal computers were considered a preferred alternative to heavy books, scrolling was identified as difficult for those with wrist pain. Thus, every effort was made to avoid this necessity. This criterion also contributed to the expansion of the overall programme in terms of number of webpages per session. In the final iteration, each module was comprised of approximately 30-40 webpages.

The literature is unclear as to the optimal dose-response relationship, particularly regarding paediatric populations. The original 20 module programme was condensed to 9 modules to be delivered weekly (see Table 5.8). The revised format for the programme was informed by similar interventions, the developmental level and limited attention span of the target sample and existing literature which supports the effectiveness of short-term self-management interventions (Carnes et al., 2012). To reduce the burden of using this complex, intervention, the structure of the *Feeling Better* intervention is intentionally streamlined to ensure participants remain focused on the intended topics. Table 5.9 and Figure 5.11 illustrate the linear structure of the final iteration of the *Feeling Better* intervention and website. This linearity is suited to tunnelled navigational options and to the pain-interference identified in user-testing. The programme needed to be designed and used as ergonomically as possible. For example, children with JIA-associated wrist pain identified scrolling as quite difficult. Ease of use was informed by the preceding qualitative study (Iteration 1) in which children offered solutions to obstacles they experience daily. In response to

qualitative feedback, a “return to where you left off” feature was embedded in the site to facilitate easy return to the last module completed and to help the programme feel less cumbersome to use (see Figure 5.9) A comparison between the manualised *Feeling Better* programme and the final iteration of the online prototype is outlined in Chapter 2 (Table 2.5). A linear structure was considered an appropriate fit for CBT which takes a guided discovery approach to the introduction strategies whereby learning is facilitated in a graded way, building in complexity as the patient progresses.

The theme of the *Feeling Better* programme was changed from a camping survivalist theme initially considered (Iteration 1) to the favoured pirate theme following early feedback from child participants. The appearance of the *Feeling Better* website needed to be appealing to children as appropriate design features may help to retain participants if a positive emotional response is associated with intervention use (Morrison et al., 2012; Yardley et al., 2015). Some suggestions for improvement in appearance and layout were not within the budget and constraints of the current study. For example, inclusion of video-based patient narratives on the application of coping skills using actual patients rather than animations. However, when asked “how much did you like the way the *Feeling Better* programme looked?” all participants answered ‘mostly’ or ‘very’ (see Tables 5.5-5.6). This suggests a degree of success in creating a visually appealing source of support for younger children which might translate into greater engagement and adherence to the programme in feasibility testing.

Table 5.8

Comparison of the manualised Feeling Better programme and the online Feeling Better intervention.

	<i>Feeling Better</i> manual for carers (McManus and McGuire, 2010)	Web-based, <i>Feeling Better</i> programme for children with chronic pain
Description	A treatment manual designed to provide practical guidance for health workers who work with people who have intellectual disabilities and chronic pain.	A pain management programme designed to teach pain coping skills for pre-adolescent children with chronic pain and their care-givers.
Reading level	Adult	Middle childhood
Content of each module:		
Weekly check-in and check out	Paper-based review of progress and practice	Interactive review of progress and practice using multiple choice questions and 'click and select' graphics
Key learning objectives	Paper-based list of objectives	List of objectives presented as treasure to be discovered
Rationale for each CBT-based technique	Text-based information	Text, video and audio-based information
Time commitment	Text-based information	Time commitment and progress bar illustrating progress through the module
Practical guidance on how to conduct the session	Text-based information	Text-based information, automated messages (instruction or encouragement); video materials with instructions, graphics illustrating use of certain techniques
Tips for more effective practice	Text-based information	Tips for more effective practice presented as clues

Case examples	Text-based information	Pirate themed, age appropriate patient narratives linked to interactive exercises
Handouts	Text-based information	Printable pdf versions of all techniques covered in the programme, released as each module is completed.
DVD	DVD	DVDs and addition video materials including animated, education-based videos and cartoons
Client summary: recap of content covered	Text-based information	Client summary in the form of a Weekly quiz
Homework assignments (paper-based)	Text-based information	Homework assignments in form of an interactive, 'final challenge', submitted online
Module structure	Module 1: Understanding pain (education)	Introduction
	Module 2: Relaxation - deep breathing	Module 1: Understanding pain (education)
	Module 3: Relaxation - progressive muscle relaxation	Module 2: Relaxation training - deep breathing
	Module 4: Relaxation - guided visualisation	Module 3: Exercise, activity pacing - and progressive muscle relaxation
	Module 5: Physical exercise	Module 4: Attention management - and taking your mind off the pain
	Module 6: Activity pacing	Module 5: Attention management - changing pain sensations

Module 7: Attention management skills - taking your mind off the pain - Module 6: Thoughts and feelings - and guided visualisation

Module 8: Attention management skills - changing pain sensations - Module 7: Problem solving

Module 9: How your thoughts make you feel - Module 8: Communication skills

Module 10: Challenging negative thoughts - Module 9: Future planning - relapse prevention

Module 11: Positive thinking, coping self-talk

Module 12: Goal setting

Module 13: Problem solving

Module 14: Positive communication

Module 15: Medication

Module 16: Relapse prevention

Module 17: Follow-up session - Review I

Module 18: Follow-up session - Review II

Module 19: Follow-up session - Review III

Module 20: Follow-up session - Review IV

Table 5.9
Structure of each module in the Feeling Better programme.

Captain's Log: weekly check-in.	The rationale, objectives and outline of the module. Re-cap of last module and homework. Addresses the question: "What will I discover (learn) in this module?" and "Why is this module important?"
Main content	Each module typically requires 30 minutes to complete. The main content webpages introduce or develop a new topic and set of CBT strategies for chronic pain management using vignettes, video and audio materials, illustrations and images, interactive exercises, questions and responses. Core points are highlighted in the text and simplified for ease of understanding.
Case example	Every module has a at least two vignette / testimonials which introduce a problem or barrier and a solution or facilitator
Your Story	Interactive, If-then planning exercise
Case example	Every session has a summary page that provides a review of the core points presented in the session.
Your Story	Interactive, If-then planning exercise
Quiz	Every module features a client summary presented in the form of an interactive quiz. This summarises core points covered in the associated module.
Homework	Eight out of nine modules feature homework specific to the theme of the session
Well done!	Positive reinforcement in the form of encouraging messages and a re-cap of what was covered in associated module.
Schedule next module	Participants receive an email reminder for their next module and progress report for the preceding week.



Figure 5.11 Layout and structure of the *Feeling Better* intervention website

5.5.4 Behavioural prescriptions (ease of understanding)

To facilitate ease of understanding the following modifications were implemented. Simplified response options e.g. multiple choice or pre-defined lists, additional pages featuring worked examples of the more difficult CBT-strategies and more reminders of how to use certain strategies and how to contact the online tutor for assistance. In Iteration 1 and 2, children opted for list-based information and multiple choice, quiz or game-like terminology and answer options to ease the cognitive demands of the programme (Iteration 1 and 2; think aloud and PRP groups). These suggestions were accommodated in Iteration 3 and 4 (survey and expert testing). The information provided by the *Feeling Better* programme was staggered and presented using a more graded style of delivery. For example, the relaxation training techniques initially presented in Module 2 were dispersed throughout the programme and SMART goal setting was introduced in Module 1, instead of Module 7 and revisited in the weekly check-in at the start of each module. Also, more graphics or visual illustrations were added in place of text-based content (see Figure 5.12 and 5.13 visual illustration of ideas).

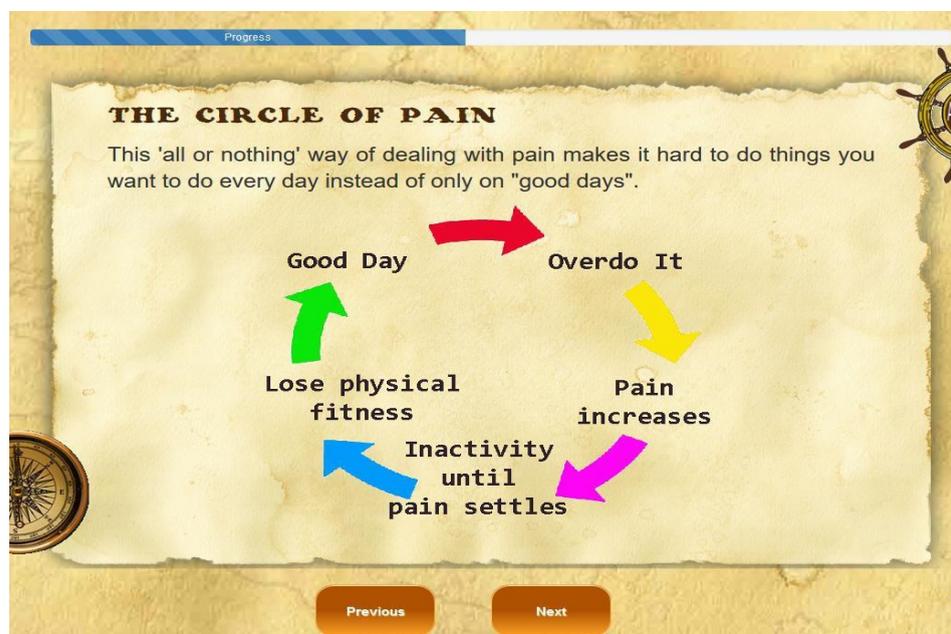


Figure 5.12 Visual illustration to ease understanding

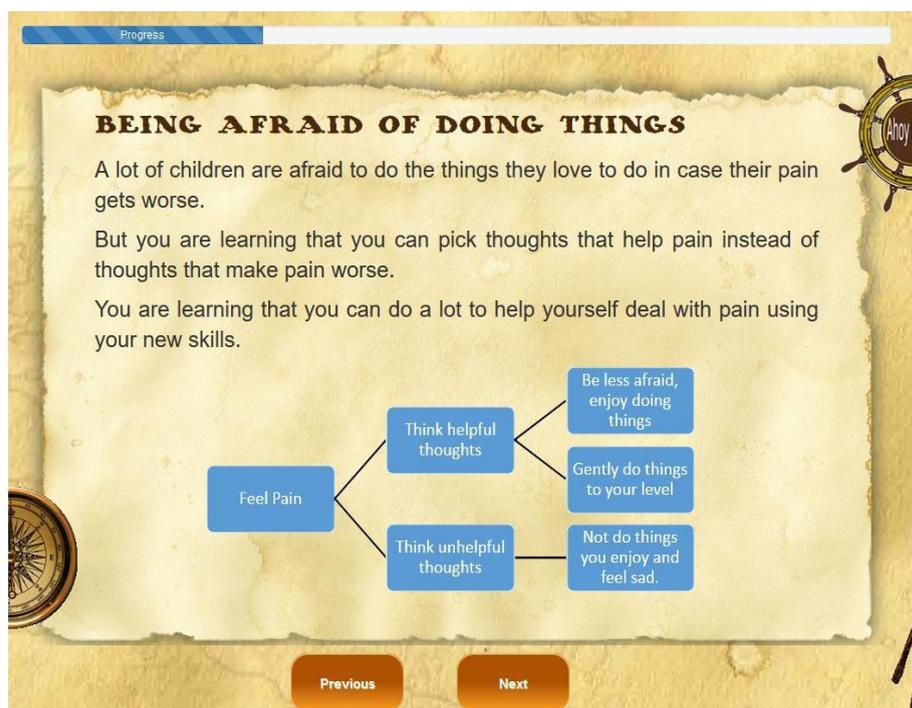


Figure 5.13 Visual illustration to present more complex concepts

Many of the negative comments collected referred to issues around understandability and readability defined in terms of behavioural prescriptions. Interactive thought challenging exercises and BEST communication plans were considered least helpful and too sophisticated for this age group: “Simplify, make it easier for her to do” (PG4). In Iteration 3, a total of child participants (Child SG4; survey group) questioned how to use the “Treasure Hunt” (i.e. thought challenging exercise) (see Figure 5.14) and others struggled to understand how to complete the BEST communication plan. In Iteration 4, one expert identified these strategies as too sophisticated (see Tables 5.6). Following this feedback, structural changes were made to these CBT-strategies to allow ease of understanding and reduce cognitive load. In addition, simplified instructions for use and a worked example was provided on the page before each exercise. The more complex CBT strategies e.g. problem-solving, communication skills and cognitive-restructuring were again revised to accommodate children at the lower end of the target age range.

Progress

FINAL CHALLENGE - TREASURE HUNT

Unhelpful / Helpful Thought

Clue: I will never be able to sleep

Evidence it is True

Clue: I couldn't sleep last night

Evidence it is False

Clue: I have been able to sleep on other nights

Belief

Medium

Walk the Plank Treasure Chest

Previous Next

Figure 5.14 Example thought challenging exercise

Consideration of emotional or cognitive limitations was further demonstrated by the range of alternative learning opportunities presented. For example, the therapeutic materials in the *Feeling Better* programme offer the child and parent (i.e. lay therapist) a choice of strategies. If the child at the younger end of the target age range is having difficulty grasping a more complex exercise, alternative and less sophisticated exercise options i.e. interactive strategies that facilitate learning and behaviour acquisition, are available. In addition, easier, alternative exercises are placed immediately following more complex techniques to facilitate e.g. move on from thought challenging exercise and attempt progressive muscle relaxation instead. In Iteration 3, when asked “how easy was the information to understand?” most children responded “somewhat” whereas the majority answered “mostly” (see Table 5.4). In addition, feedback from Iteration 4 suggest further copy-editing of the text-based clinical content was required. Therefore, a page by page revision of all text in the intervention website was conducted and text-based content simplified prior to further feasibility testing. Blank copies of all core CBT-strategies were automatically saved in

the “Pirate Pouch” upon completion of the given skill. This feature was designed to encourage engagement with the programme and off-line practice of certain strategies using the printable pdfs provided.

Finally, pre-adolescent children are particularly attuned to learning through modelling and imitation and are highly influenced by parenting behaviour (Bandura, 1996). Therefore, patient case examples were littered throughout each module of the *Feeling Better* programme in both the parent and child sections (see Figure 5.15). Observational learning was further supported using video materials. This strategy is implemented in a manner consistent with the original FB manual. However, to address parameters for effectiveness, the patients in the patient narratives were presented as children, the content of each narrative was reduced, refined and made relevant to the target population. Parent case examples were tailored to present problems pertinent to parenting behaviour.

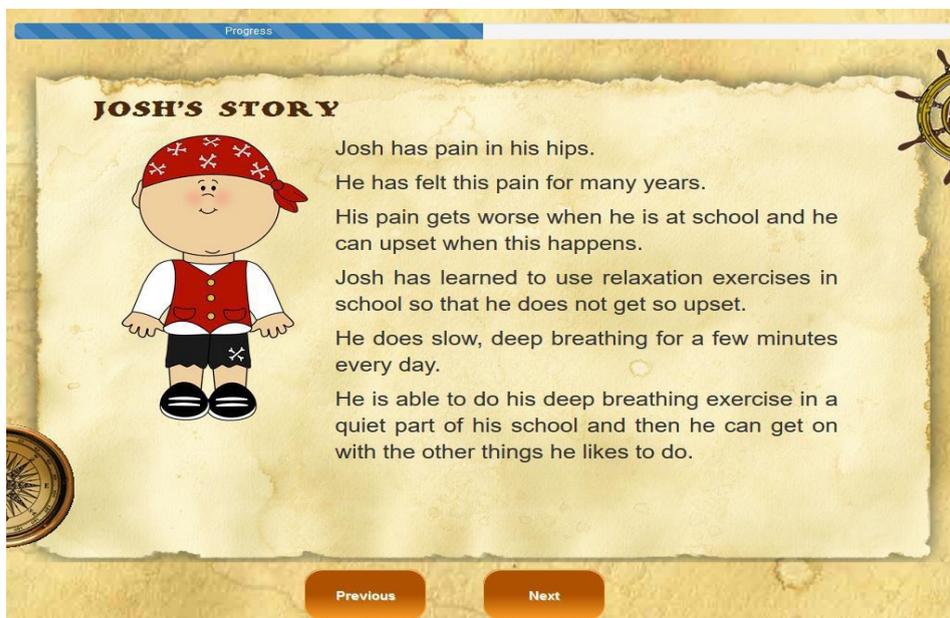


Figure 5.15 Example of the modified patient narrative

5.5.5 Support (social)

A secure encrypted internal messaging system was incorporated in the intervention website. This enabled two-way communication between the

online tutor and participants. Child and parent participants could ask questions and receive answers relating to the use of certain strategies, difficulties experienced and progress so far. The function of this feature was to provide support and to motivate. Supported or guided pain self-management in addition to clinical content differentiates this intervention from information-only alternatives. It is thought the provision of supplementary support might contribute to intervention engagement and retention (Morrison et al., 2014).

Most of the negative comments collected from children and parents were mapped to this domain. Parents and children agreed on the need for human support and most participants opted for communication with experts and peers in addition to an online tutor. Most participants thought the “inclusion of a figure (e.g. a virtual coach) to demonstrate the exercises would be helpful” (Parent SG4). For example, “provide access to experts” (n= 6, PG2); “can we share this pain information with her doctor” (n=8, PG3); “like to talk to other children using the programme” (n=4, CG3; n= 8, PG3). These features i.e. social support and context: peer to peer contact or simulated person to person interaction are associated with positive outcomes and more effective interventions (Morrison et al., 2012; Morrison, 2015). However, their implementation was beyond the budget and resources available to this research. Some preferences were accommodated. For example, the provision of information about others i.e. vignettes/social norms and social support from the online tutor, personalised messages of encouragement and progress reports from an online tutor via email (parents) and internal messaging system (children).

Children received automatic encouragement in response to interaction with the website e.g. automated messages. This facility is associated with more effective online interventions (Palermo et al., 2009; Morrison et al., 2012; Palermo et al., 2016). All parent participants reported being happy with the features that facilitate personalised correspondence with the online tutor i.e. weekly progress reports, internal messaging. Older children were comfortable using the internal message centre (*Pirate Post*)

but younger children were reluctant to use it (Iteration 3; parent and child survey groups). In Iteration 4 an instant messaging application was incorporated in the programme, embedded in the home page for use by participants and non-participants (pre- registration, general enquiries). Participants preferred guided rather than self-led progression through the programme but suggested the online tutor be referred to as a “coach” to make the programme less like school-like. Following initial registration and enrolment, only those parents who experienced issues with technology or access to the internet opted to use the telephone to speak with the project co-coordinator.

5.5.6 Content (CBT; accurate, clear, simple)

The clinical content of this intervention is entirely derived from the *Feeling Better* manual (McManus & McGuire, 2010). As such it is based on CBT principles. The need to address pain-related cognition and affect was established in Study 2 and facilitated in each Iteration of intervention development (Iteration 1 & 3). Parent participants described the need to address cognitive biases such as fear avoidance, the belief that pain indicates damage, as well as the depression, anxiety and stigma related to the use of psychological therapy for pain management support. Self-efficacy, outcome expectations, behaviour regulation, goals and facilitation were identified as important determinants of pain behaviour and behaviour change. The main constructs of SCT were implemented at a practical level using CBT-based strategies believed to address these mechanisms of change e.g. SMART goals (Behavioural goals), video demonstrations of behaviour (observational learning: vicarious learning/modelling), weekly check-in and check-out (self-monitoring), activity pacing (Behaviour regulation). Also, the provision of social rewards in the form of virtual promotion up through the ranks as part of pirate theme (incentive motivation) and weekly progress reports in which parent participants received feedback on their child’s progress through the programme thus far (e.g. positive reinforcement) and

reminders about homework activities and access to the online tutor (Behavioural capability). In turn, children were supported through an internal email system which allowed them to communication with the online tutor (social support). Patient narratives were each followed by a ‘*Your Story*’ page that afforded an opportunity for personal reflection on how the case example might apply to the child participant (behaviour regulation) (see Figure 5.16).

Figure 5.16 Example of the Your Story exercise

Check-in and check-out features functioned as reminders in weekly progress report and as opportunities for self-monitoring and self-reflection (Behaviour regulation). Pain education (behavioural capability), goals, rewards, progress reports (behavioural goals), self-monitoring and opportunities for accomplishment (i.e. mastery experience) were incorporated to address participants perceived competence for coping (i.e. self-efficacy) and motivation to engage with the *Feeling Better* programme. Demonstration of behaviour and opportunities for observational learning are particularly important strategies for this target group. Further, research is needed to determine if outcomes could be improved by use of different, relevant role models (Morrison et al., 2012). BCTs (e.g. goal setting, prompts, rehearsal) and MoDs (e.g. interactive exercises, quizzes) were

applied accordingly.

5.5.7 Delivery

A variety of concrete learning tools were used e.g. supporting text with video and audio materials and graphic illustrations. Video and audio materials either demonstrated techniques or presented explanation and instruction for task performance. A quiz was presented at the close of 8 treatment sessions. These quizzes functioned as client summary sheets, recapping the lessons learned and de-briefing the participant. There were approximately 5 or more interactive exercises per treatment session. User preferences for modes of delivery were mixed. Differences emerged on the delivery of patient narrative using video recordings of real patient testimonials (this was beyond the scope of this study) or an online character i.e. avatar or static character: The online *Feeling Better* programme has animated, and static illustrations embedded in the website. There are approximately two vignettes per module and each feature a graphic of a child, consistent with the pirate themes. Child group preferred the use of a graphic character to illustrate patient narratives and CG2 and CG3 opted for a mixture of 2D vignettes and video recordings of real-time, patient testimonials. The resources for this project did not stretch to include the latter.

5.5.8 Assessment

It was decided at the outset that tailoring according to individual user needs and preferences was beyond the remit and resources supporting this research. Therefore, while the *Feeling Better* programme was personalized throughout, the clinical content and self-management recommendations were not tailored to meet individual needs (see Figure 5.17). Research exists to support both tailored and non- tailored treatment delivery (Noar, Benac, & Harris, 2007; Morrison et al., 2012). Given that this is an exploratory

study our aim was to design a programme that was all-encompassing. Future iterations of the programme may explore or compare the acceptability of both approaches in this cohort.

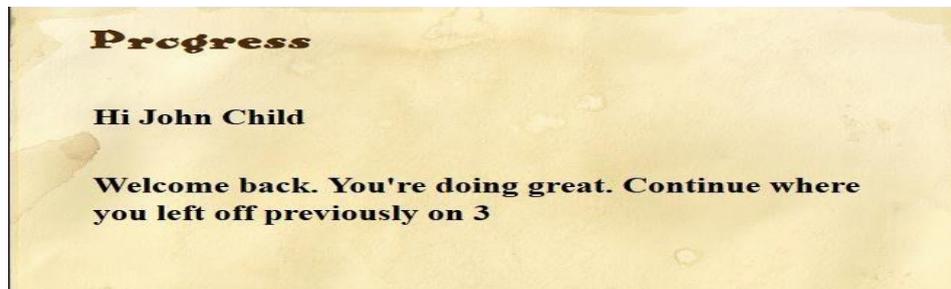


Figure 5.17 Example personalised message encouraging progress

5.5.9 Burdens (use of content not technology)

The literature offers relatively little guidance on how best to adapt and implement established clinical content (*Feeling Better* manual). Particularly for the reading level and developmental capabilities of relatively wide-ranging pre-adolescent age group (5-12 years). For ease of understanding visual illustrations of key concepts were provided where it was possible to present the idea or issue in graphic form. Research relating to the sensitivity of CBT for younger children suggests visual illustration of key concepts is optimal for the age group targeted (Nelson & Tusaie, 2011; Tusaie, 2011). This approach is consistent with website design principles that the use of short, concise texts which present one idea per paragraph and make use of supplemental options such as interactive features and visual or audio materials (van Merriënboer & Sweller, 2010). To foster comfort when completing an emotion-focused strategy, the child was treated as the “expert” and asked to offer advice (or create supported/ guided coping statements) for their own use or that of other children who might have pain like theirs i.e. write their experiences in a way that diffuses the emotional content and empowers the child.

5.5.10 The online *Feeling Better* programme – Intervention components

Modifications to text-based instruction on how to perform behaviours were necessary to simplify intervention content in terms of intensity and ease of understanding. This process influenced the coding applied to some BCTs which were previously coded as present beyond all reasonable doubt and subsequently coded as ‘present in all probability’ by two independent researchers (AT, EM) (Table 5.10). The content of each of the *Feeling Better* treatment sessions were analysed independently by two coders, one of whom was unfamiliar with the intervention. Disagreements between the two reviewers were resolved by discussion until consensus was reached. Table 5.10 shows the inter-rater reliability for each of the *Feeling Better* treatment modules. According to the guidelines from Landis and Koch (1977), the strength of agreement between the reviewers was moderate across modules and overall (average $\kappa = .618$, $p < .05$).

Table 5.10
Inter-rater reliability of BCT coding for each module

Modules	κ
Introduction	1.000
Module 1	1.000
Module 2	0.552
Module 3	0.557
Module 4	0.552
Module 5	0.442
Module 6	0.557
Module 7	0.503
Module 8	0.451
Module 9	0.564
Average	0.618

In line with the therapeutic process (i.e. CBT), the clinical content of each successive FB treatment sessions becomes more complex. Each treatment session is designed to build on the lessons learned in the previous and to promote practice of previously acquired skills as well as those

introduced in each weekly session. For example, guided practice of progressive muscle relaxation following an introduction to cognitive restructuring. The number of BCTs did not however increase with each session, simply the complexity of the CBT strategy employed. The structure of each treatment session was deliberately consistent with the previous. This was designed to decrease the cognitive demand placed on younger children and promote ease of use and understanding. The treatment session with the most BCTs was treatment session 6. The most used BCTs were 4.1. Instruction on how to perform the behaviour; 1.1 Goal setting (behaviour); 1.3 Goal setting (outcome); Demonstration of the behaviour; These BCTs featured in every treatment session.

Table 5.11 presents a component analysis of the final version of the *Feeling Better* website which outlines the intervention components incorporated in the final draft of the website. CBT for chronic pain is comprised of the following components: education, goal setting, relaxation, graded activity, operant principles, behavioural experiments, attention management, cognitive restructuring, problem solving and maintenance of active coping behaviour. Strategies from each of these domains were incorporated in the *Feeling Better* intervention. This was facilitated using a total of 23 different BCTs. The most common of which were “goals (behaviour) and goals (outcome)”, “activity planning”, “instruction on how to perform behaviour”, “information about health consequences” and “demonstration of behaviour”.

The final version of the online *Feeling Better* intervention features 13 different modes of delivery as outlined in the classification scheme proposed by Webb et al (2012) (see Table 5.12). These design features support the application of the outlined BCTs and CBT-strategies that comprise this intervention. All automated and communicative functions were present in each treatment session.

Table 5.11

Feeling Better Intervention Components

Module	Content/Strategy	SCT Construct	BCT	Mode of delivery
Introduction	Rationale; What is CBT; What to expect; Commitment to training	Knowledge	5.1. Information about health consequences; 5.3. Information about social and environmental consequences; 5.6. Information about emotional consequences; 9.1. Credible source; 12.5. Adding objects to the environment;	Information about chronic pain, coping skills and self-management; Video: “What is Chronic Pain”; Handouts
Module 1 Pain Education	Provide information about behaviour-health link; Illustrate interaction between thoughts and feelings; Activity monitoring; SMART Goal Setting; Graded tasks; Barrier / facilitator identification	Self-efficacy; Pain beliefs (outcome expectations, pain catastrophising); behavioural goals; facilitation; Impediments	1.1. Goal setting (behaviour); 1.2. Problem solving; 1.3. Goal setting (outcome); 1.4. Action planning; 1.6. Discrepancy between current behaviour and goal; 2.4. Self-monitoring of outcome(s) of behaviour; 3.1. Social support (unspecified); 4.1. Instruction on how to perform the behaviour; 4.2. Information about antecedents; 5.1. Information about health consequences; 6.1. Demonstration of the behaviour; 8.1. Behavioural practice/rehearsal; 8.2. Behaviour substitution; 8.3. Habit formation; 15.1. Verbal persuasion about capability; 16.2. Imaginary reward	Interactive online lesson, Video, Quiz, Homework. Content: Information about chronic pain, coping skills and self-management; Video: “What is Chronic Pain”; Handouts, Peer-based patient narratives.

Table 5.11
Feeling Better Intervention Components (continued...)

Module	Content/Strategy	SCT Construct	BCT	Mode of delivery
Module 2 Relaxation training	Review of progress; Activity monitoring; SMART Goal Setting; Graded tasks; Relaxation training; Identify barriers / facilitators	Self-efficacy; Pain beliefs (outcome expectations, pain catastrophising); behavioural goals; facilitation; Impediments	1.1. Goal setting (behaviour); 1.2. Problem solving; 1.3. Goal setting (outcome); 1.4. Action planning; 1.6. Discrepancy between current behaviour and goal; 2.4. Self-monitoring of outcome(s) of behaviour; 3.1. Social support (unspecified); 4.1. Instruction on how to perform the behaviour; 4.2. Information about antecedents; 5.1. Information about health consequences; 6.1. Demonstration of the behaviour; 8.1. Behavioural practice/rehearsal; 8.2. Behaviour substitution; 8.3. Habit formation; 11.2. Reduce negative emotions; 15.1. Verbal persuasion about capability; 16.2. Imaginary reward	Interactive online lesson, Video, Quiz, Homework. Content: Information about relaxation and physiological sensations, coping skills and self-management; Video: “Deep breathing”; Handouts, Peer-based patient narratives. Change objectives: Changing expectations about the utility of coping skills

Module 3 Exercise & activity pacing	Review of progress; Activity monitoring; SMART Goal Setting; Graded tasks; Guided discovery of behaviour patterns; Activity planning; Alternating activity; Relaxation training;	Self-efficacy; Pain beliefs (outcome expectations, pain catastrophising); behavioural goals; facilitation; Impediments	1.1. Goal setting (behaviour); 1.2. Problem solving; 1.3. Goal setting (outcome); 1.4. Action planning; 1.6. Discrepancy between current behaviour and goal; 2.4. Self-monitoring of outcome(s) of behaviour; 3.1. Social support (unspecified); 4.1. Instruction on how to perform the behaviour; 4.2. Information about antecedents; 5.1. Information about health consequences; 6.1. Demonstration of the behaviour; 8.1. Behavioural practice/rehearsal; 8.2. Behaviour substitution; 8.4. Habit reversal; 15.1. Verbal persuasion about capability;	Interactive online lesson, Video, Quiz, Homework. Content: Information about exercise and activity pacing, coping skills and self- management; Video: Progressive muscle relaxation; Handouts, Peer-based patient narratives. Change objectives:
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Table 5.11

Feeling Better Intervention Components (continued...)

Module	Content/Strategy	SCT Construct	BCT	Mode of delivery
Module 4 Attention management - focusing	Review of progress; Activity monitoring; SMART Goal Setting; Relaxation training; Barrier / facilitator identification;	Self-efficacy; Pain beliefs (outcome expectations, pain catastrophising); behavioural goals; facilitation; Impediments	1.1. Goal setting (behaviour); 1.2. Problem solving; 1.3. Goal setting (outcome); 1.4. Action planning; 1.6. Discrepancy between current behaviour and goal; 2.4. Self-monitoring of outcome(s) of behaviour; 3.1. Social support (unspecified); 4.1. Instruction on how to perform the behaviour; 4.2. Information about antecedents; 5.1. Information about health consequences; 5.1. Information about health consequences; 5.4. Monitoring of emotional consequences; 6.1. Demonstration of the behaviour; 8.1. Behavioural practice/rehearsal; 8.2. Behaviour substitution; 8.4. Habit reversal; 11.2. Reduce negative emotions; 12.4. Distraction; 15.1. Verbal persuasion about capability; 16.2. Imaginary reward	Interactive online lesson, Video, Quiz, Homework. Content: Information about attention management, coping skills and self-management; Video: “Guided visualisation”; Handouts, Peer-based patient narratives. Change objectives: Changing expectations about the utility of coping skills

Table 5.11

Feeling Better Intervention Components (continued...)

Module	Content/Strategy	SCT Construct	BCT	Mode of delivery
Module 5 Attention management- changing sensations	Review of progress; Activity monitoring; SMART Goal Setting; Relaxation training; Barrier (and facilitator) identification;	Self-efficacy; Pain beliefs (outcome expectations, pain catastrophising); behavioural goals; facilitation; Impediments	1.1. Goal setting (behaviour); 1.2. Problem solving; 1.3. Goal setting (outcome); 1.4. Action planning; 1.6. Discrepancy between current behaviour and goal; 2.4. Self-monitoring of outcome(s) of behaviour; 3.1. Social support (unspecified); 4.1. Instruction on how to perform the behaviour; 4.2. Information about antecedents; 5.1. Information about health consequences; 5.1. Information about health consequences; 5.4. Monitoring of emotional consequences; 6.1. Demonstration of the behaviour; 8.1. Behavioural practice/rehearsal; 8.2. Behaviour substitution; 8.3. Habit formation; 11.2. Reduce negative emotions; 12.4. Distraction; 15.1. Verbal persuasion about capability; 16.2. Imaginary reward	Interactive online lesson, Video, Quiz, Homework. Content: Information about attention management, coping skills and self-management; Video: “Attention focusing”; Handouts, Peer-based patient narratives.

Table 5.11

Feeling Better Intervention Components (continued...)

Module	Content/Strategy	SCT Construct	BCT	Mode of delivery
Module 6 Thoughts & Feelings	Review of progress; Activity monitoring; SMART Goal Setting Cognitive reappraisal; Thought records; Thought challenging; Relaxation training;	Self-efficacy; Pain beliefs (outcome expectations, pain catastrophising); behavioural goals; facilitation; Impediments	1.1. Goal setting (behaviour); 1.2. Problem solving; 1.3. Goal setting (outcome); 1.4. Action planning; 1.6. Discrepancy between current behaviour and goal; 2.4. Self-monitoring of outcome(s) of behaviour; 3.1. Social support (unspecified); 4.1. Instruction on how to perform the behaviour; 4.2. Information about antecedents; 5.1. Information about health consequences; 5.4. Monitoring of emotional consequences; 5.6. Information about emotional consequences; 6.1. Demonstration of the behaviour; 8.1. Behavioural practice/rehearsal; 8.2. Behaviour substitution; 8.3. Habit formation; 11.2. Reduce negative emotions; 13.2. Framing/reframing; 13.3. Incompatible beliefs; 15.1. Verbal persuasion about capability;	Interactive online lesson, Video, Quiz, Homework. Content: Information about thoughts, feelings and behaviour, coping skills and self-management; Video: “Guided visualisation”; Handouts, Peer-based patient narratives.

Module 7 Problem solving	Review of progress; SMART Goal Setting; Problem solving e.g. BEST acronym; Relaxation skills; Relaxation training;	Self-efficacy; Pain beliefs (outcome expectations, pain catastrophising); behavioural goals; facilitation; Impediments	1.1. Goal setting (behaviour); 1.2. Problem solving; 1.3. Goal setting (outcome); 1.4. Action planning; 1.6. Discrepancy between current behaviour and goal; 2.4. Self-monitoring of outcome(s) of behaviour; 3.1. Social support (unspecified); 4.1. Instruction on how to perform the behaviour; 4.2. Information about antecedents; 5.1. Information about health consequences; 6.1. Demonstration of the behaviour; 8.1. Behavioural practice/rehearsal; 8.2. Behaviour substitution; 8.3. Habit formation; 15.1. Verbal persuasion about capability;	Interactive online lesson, Video, Quiz, Homework. Content: Information about problem solving, coping skills and self-management; Video: “Deep breathing”, Peer-based patient narratives. Change objectives: Changing expectations about the utility of coping skills
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Table 5.11

Feeling Better Intervention Components (continued...)

Module	Content/Strategy	SCT Construct	BCT	Mode of delivery
Module 8 Communication skills	Review of progress; SMART Goal Setting; Relaxation training; Barrier / facilitator identification;	Self-efficacy; Pain beliefs (outcome expectations, pain catastrophising); behavioural goals; facilitation; Impediments	1.1. Goal setting (behaviour); 1.2. Problem solving; 1.3. Goal setting (outcome); 1.4. Action planning; 2.4. Self-monitoring of outcome(s) of behaviour; 3.1. Social support (unspecified); 4.1. Instruction on how to perform the behaviour; 4.2. Information about antecedents; 5.1. Information about health consequences; 6.1. Demonstration of the behaviour; 8.1. Behavioural practice/rehearsal; 8.2. Behaviour substitution; 8.3. Habit formation; 15.1. Verbal persuasion about capability;	Interactive online lesson, Video, Quiz, Homework. Content: Information about communication skills, coping skills and self-management; Video: “Progressive muscle relaxation”; Handouts, Peer-based patient narratives.
Module 9 Future planning	Relapse prevention training;	Self-efficacy; Pain beliefs (outcome expectations, pain catastrophising); facilitation; Impediments	1.1. Goal setting (behaviour); 1.2. Problem solving; 1.3. Goal setting (outcome); 1.4. Action planning; 1.6. Discrepancy between current behaviour and goal; 2.4. Self-monitoring of outcome(s) of behaviour; 3.1. Social support (unspecified); 4.1. Instruction on how to perform the behaviour; 4.2. Information about antecedents; 5.1. Information about health consequences; 6.1. Demonstration of the behaviour; 8.1. Behavioural practice/rehearsal; 8.2. Behaviour substitution; 8.3. Habit formation; 15.1. Verbal persuasion about capability	Interactive online lesson, Video, Quiz, Homework. Content: Information about future planning, coping skills and self-management; Handouts, Peer-based patient narratives.

Table 5.12

Feeling Better Intervention: Mode of delivery analysis

Mode of delivery: Automated functions (tick all that apply)	Child Pathway	Parent Pathway
(a) Enriched information environment (e.g. supplementary content and links,	✓	✓
(b) Automated tailored feedback based on individual progress monitoring (e.g. comparison to goals, reinforcing messages, coping messages etc.)	✓	✓
(c) Generic follow-up messages (e.g. reminders, tips, newsletters, encouragement)	✓	✓
(d) Tailored initial advice (on basis of answers to questions about beliefs, problems, circumstances).		
(e) Supported progress monitoring (e.g. electronic diary/chart).	✓	✓
Mode of delivery: Communicative functions (tick all that apply)		
(f) Access to advisor to request advice (e.g. 'ask the expert' facility, expert-led discussion board or chat sessions)	✓	✓
(g) Scheduled contact with advisor (e.g. emails)	✓	✓
(h) Peer-to-peer access (e.g. buddy systems, peer-to-peer discussions boards/forums, live chat)		
Supplementary modes of delivery:		
(i) Email	✓	✓
(j) Telephone	✓	✓
(k) Text message (SMS)		
(l) CD ROM		
(m) video conferencing		
(n) Web-based		
(o) Other (specify): Instant-messaging via Live Chat application	✓	✓
Navigational format:		
(p) tunnelled (specify): progress dependent upon completion of preceding module	✓	✓
(q) free choice (specify)		
Entertainment value:		
(r) quizzes, stories, graphics (specify)	✓	✓
(s) appearance (e.g. colour, layout etc.) (specify)	✓	✓
Credibility:		
(t) sources, credentials cited (specify)	✓	✓
(u) other specify		

5.6 Discussion

5.6.1 Overall findings

The online *Feeling Better* programme was developed following a comprehensive assessment of intervention components in line with Ritterband's Behaviour Change Model for Internet Interventions (2009). This study was conducted to identify issues with function, relevance and ease of understanding. Usability testing showed the online *Feeling Better* programme was considered acceptable, relevant, easy to use and understand for users in the target age group.

5.6.2 Usability and functionality

Mixed methods evaluation of usability was considered successful. Had we relied on one method our understanding of usage (i.e. individual interaction with the platform) and functionality (i.e. technical issues) might have been incomplete. This research identified usability issues that if not corrected or modified, could have diminished any potential benefit that may be derived from the online *Feeling Better* programme. For example, a recurring issue was the sophistication of the language used. Later iterations identified difficulty when using the thought challenging, communication and problem-solving techniques. Ideas and issues were identified in each iteration that were important to address prior to a larger trial.

Online testing identified functionality issues e.g. optimal web-browsers, screen-size that affected operational website features. For example, participant response to open-ended questions identified functionality issues in relation to tablet screen size and web browser use whereby the progress bar could not be viewed on tablets of less than 17 inches and the programme failed to operate on older version of the Internet Explorer web-browser. The programme was subsequently optimized for a smaller screen size and recommendations for optimal use could be made to all eligible participants. The end-product was an intervention ready to be evaluated by pre-adolescent children with chronic pain and their care-givers

in a feasibility trial, the methods and results of which are presented in the following chapter. Loss of eligible participants due to technical or functionality errors could have had an enormous impact on the conduct of the *Feeling Better* trial. Had we relied on feedback from children or parents only, we may not have captured the various issues about ease of understanding and readability which might have been a barrier to use.

5.6.3 Website design preferences

A list of preferred design features and techniques was created that contributes to a currently limited understanding of the website elements that appeal to younger children with chronic pain. These findings suggest interventions that prioritise individual goals, progress and feedback on behaviour, and contain personalised information are more likely to engage younger children with chronic pain.

Differences and similarities were observed between child and parent users in terms of their preferred website design features. Child and parent preferences differed in terms of the extent of the delivery of patient narratives e.g. parents preferred real patient case examples delivered using video recording, whereas children preferred the use of animated game-like characters to demonstrate behaviour.

Child and parent preferences in relation to the nature and extent of parent involvement were also mixed. Younger children (9 years or lower) preferred to complete the programme in collaboration with their mother whereas older children requested personal profiles, their own password and generally more independent use of the online system.

User feedback from the think-aloud and PRP workshops indicated user preference for clear, simple instructions, relatively little text per page, visual illustration of concepts and repetition of key concepts. These preferences were expressed by most children and parents regardless of literacy level. The general preference is consistent with cognitive load theory which suggests that reducing the cognitive demand placed on the individual will promote optimal learning (Doak, Doak & Root, 1996).

Children and parents opted for a tunnelled navigation rather than free choice. Guided navigational procedures were considered more appealing and less-distracting from the purpose of each treatment session. Tunnelled navigation made the *Feeling Better* programme seem easier to use, possibly because this approach reduces the cognitive demand placed on the user engaging with the programme.

Both children and parents reported a preference for a social network which offers access to peers and experts. Most of the negative comments referred to the lack of this design feature. These preferences validate the findings from Study 2 referring to barriers and facilitators of pain self-management which suggest further work is required on communication functions to increase acceptability.

5.6.4 Usability studies

Qualitative and quantitative feedback was comparable with other usability studies focusing on chronic pain, JIA and Diabetes (Stinson et al., 2008; Long & Palermo, 2009; Stinson et al., 2010b; Nelson et al., 2016). Stinson and colleagues (2010b) explored the usability of an online programme for young people (12-18 years) with JIA. In line with the current study, suggestions for improvement were collected and children were more comfortable using the programme compared to their parents. Interactive features e.g. discussion board, inspirational stories and video recordings of JIA patients were associated with a positive-affective response.

5.6.5 Adaptation and development

This usability study builds on the findings of the preceding studies by illustrating how the *Feeling Better* manual was adapted and implemented in an online platform in a way that is consistent with person-based intervention development guidelines (Yardley et al., 2015). It should be noted, the current intervention referred to empirical evidence and theory in addition to end-user feedback because a reliance on the latter may not have

captured an accurate account of user needs. Research evaluating adult user preferences according to age have observed that while most user preferences matched the current interface user guidelines, some differences emerged. A minority of computer users reported user preferences for function and design that were not entirely based on functional (cognitive) capabilities indicating that factors such as aesthetics play an important role (Slegers, Boxtel, & Jolles, 2005). This is important to note as it is expected extended physical use of a computer will cause fatigue in some children with JIA and EDS. Therefore, despite the creativity of some participant ideas it is important to distinguish and balance user's needs with user preferences when determining intervention intensity and the components that might be incorporated.

Recommendations were not followed in some circumstances. For example, the protocol for the *Feeling Better* programme was based on group preferences rather than individual needs. Empirical evidence (Study 1), user needs and preferences (Study 2), guidelines for intervention development and the context in which this research is undertaken (i.e. the resources available to this project) were not always compatible. The challenge of successfully integrating all sources of evidence led to the development of a complex intervention which was arguably too comprehensive to tailor clinical content according to baseline assessment. Tailoring (i.e. clinical content delivered according to baseline assessment data) has been shown decrease the cognitive load placed on the user, allowing attention to focus on key topics (DiClemente, Marinilli, Singh, & Bellino, 2001). It may be that this is the optimal approach to treatment delivery for pre-adolescent children. Future research might compare the value of this feature for age-specific pain populations.

On this note, several features have been incorporated that have been listed in the literature as associated with less effective interventions or mixed findings following treatment. These include the use of avatars, additional correspondence containing usage promotion techniques and the provision of information about others (Morrison et al., 2012; Morrison et al., 2014a).

The reasons for inclusion in the *Feeling Better* programme are the feedback from participants, the resources available to the current research and the paucity of studies that illustrate preferred design features from the perspective of pre-adolescent children.

5.6.6 Strengths

A strength of this study is the approach to usability testing. A mixed methods approach was taken to develop the online programme and understand the challenges associated with its use. This was conducted across several cycles of usability testing in line with best practice recommendations (Craig et al., 2008). Usability testing literature also suggests one method of testing is inadequate to capture all usage problems (Nielsen & Mack, 1994; Fernandez et al., 2011). Functional and acceptability issues were identified and suggestions for improvement were meaningfully applied where feasible to shape each subsequent iteration of the intervention.

A second strength of this study is the person-based approach to intervention development. The novelty of this approach is that the target group were involved in each phase of intervention development from the outset of the design and development process. This is particularly useful approach given that children with chronic conditions in this age range are relatively neglected in CBT outcome studies and in pain management literature. A combination of top-down (theory-based) and bottom up (person-based) approach to intervention development has been shown to enhance the acceptability of digital interventions across patient populations (Stinson et al., 2008a; Long & Palermo, 2009; Stinson et al., 2010b; Whittemore, Grey, & Jeon, 2012; Vuorimaa & Stinson, 2013). A reliance on either approach would provide an incomplete account of user needs and preferences.

Third, a purpose-built website was used to host the adapted *Feeling Better* programme. This meant greater freedom to adapt the interface and implement interactive strategies. These freedoms were important to engage

the target population which was becoming increasingly difficult to access via recruitment in the community and via social media. Several of the design features associated with effective e-Health interventions have been incorporated in the *Feeling Better* website. A focus on the treatment delivery platform is justified given the age of the target population and the potential mediating impact of by mechanisms of change within the intervention and by use of the intervention (Ritterband et al., 2009).

Fourth, the use of CBT for chronic pain management is consistent with best practice guidelines which recommend the use of treatment with the strongest evidence base (SIGN, 2013). Moreover, the study involved potential end-users in all phases of the development process with the aim of improving the acceptability of the intervention and preventing high attrition observed in similar interventions (Fisher et al., 2015).

5.6.7 Limitations

This study has several limitations. We recruited our sample from parent-led support groups for families coping with JIA and EDS via social media. All of whom were proactive on social media and in seeking help to deal with their child's chronic pain conditions. Participants self-selected and therefore, the sample may be biased, and these findings may not be representative of the population or generalise to other patient populations. Further research is necessary to determine how this intervention might be received by a clinical patient population. It may be that the recruitment method, a circular sent to members in parent-led networks via social media, appealed only to end-users who are active on social media or already motivated to learn self-management skills. A disadvantage of this approach is that we cannot determine how many families viewed the promotion material and opted not to take part. The current self-selected group of participants seemed highly motivated. Eight (67%) of the twelve parent-child dyads who initially expressed an interest in taking part in remote user-testing were willing to participate. However due to the small number of parent-child dyads who took part in online user-testing, adherence cannot be

compared with similar interventions (Stinson et al., 2010a).

The staggered release of intervention modules (i.e. approximately two modules were evaluated at any one time) might have contributed to high participant motivation i.e. prevented assessment fatigue. However, it is a limitation of the current study that the complete programme could not be evaluated in the same way it is intended to be used i.e. weekly testing of all eight modules. Online usability testing was hampered by the time-consuming nature of the website development. It would have been preferable to have explored participant perceptions of programme complexity and intensity as this was an issue of concern given the age of child participants.

The sample was relatively small. Although research suggests at least five participants in each cycle of development is enough to identify most usability problems (Gustafson et al., 2012). The current sample size did not allow further analysis of usability according to participant characteristics (age, gender), condition (severity, duration) or experience related factors (confidence using computers). It is unknown how these factors may impact usability assessment and further research is necessary in this regard.

While it was possible to track participants logging into and out of the prototype programme (PG4; CG4), it was not possible to review usage within the website i.e. pages accessed could be recorded at this time. This is a feature to be added to the online platform prior to the feasibility trial. It will be useful to have real-time data collection of user interaction with the website to determine the best way in which to collect and organise this data.

Another limitation is the reliance on retrospective self-report of programme usability. Participant report of usability via the online survey, based on remote user-testing was submitted within a period ranging from 7 to 66 days post-use. In contrast, while the participative workshops overcame the issue of retrospective report, they are limited by the environment in which data was collected. It may be that participants were uncomfortable expressing negative opinions in front of the researchers, although this is

unlikely given the number of suggestions for improvement offered by participants. Finally, the current user feedback is focused on the online *Feeling Better* programme and may not generalise to other online interventions.

5.6.8 Conclusion

There is a dearth of studies that explore the acceptability of Internet interventions in this specific age cohort. Improving our understanding of the design features most likely to engage this population and promote optimal outcomes might enhance any potential benefit that may be derived from the use of the *Feeling Better* intervention. As demonstrated in Study 1 the literature can benefit from more explicit report of intervention development and implementation. Contributing to the current understanding of Internet interventions can help researchers and healthcare professionals to determine if, when and how these interventions might be most effective. This chapter described the development and results of usability testing of the adapted *Feeling Better* programme. This study provides initial support for the online version of the *Feeling Better* programme. The intervention was considered user - friendly, relevant and acceptable as a means of pain management support among pre-adolescent children with various types of JIA and EDS. Important issues were identified and addressed prior to conducting a controlled trial to determine the feasibility of the programme for pre-adolescent children with chronic pain. Usability testing is an essential step in ensuring the online version of the *Feeling Better* programme is acceptable and feasible and the findings were duly used to iteratively refine the prototype. It is expected that the current effort to personalise the programme according to user needs and preferences will contribute to a lower attrition rate in the planned feasibility trial.

Chapter 6 A feasibility randomised controlled trial of an online pain management programme for pre-adolescent children with chronic pain (Study 4).

6.1 Chapter overview and rationale

This chapter describes the design and conduct of a 2-armed randomized controlled feasibility trial of the online *Feeling Better* intervention that aims to improve self-efficacy for pain self-management and participant use of active coping strategies for pain management. According to Arain, Campbell, Cooper and Lancaster (2010) a pilot study is “a version of the main study that is run in miniature to test whether the components of the main study can all work together”, whereas “a feasibility study is a small study used to estimate important parameters that are needed to design the main study, such as standard deviation of outcome measures, recruitment and follow up rates” (Arain, Campbell, Cooper, & Lancaster, 2010, p. 5). The *Feeling Better* trial was designed to explore both feasibility and piloting, as a focus on both is required to inform progression to a definitive, full-scale randomised controlled trial.

6.2 Introduction

The findings from Study 3 indicated strong initial support for the usability of the online *Feeling Better* programme. Findings from this study informed each iteration of the website development and the adaption of clinical content derived from the *Feeling Better* manual (McManus & McGuire, 2010). The findings from Study 3 highlighted functional errors and issues relating to ease of understanding and relevance. With respect to web applications, there is a wealth of empirical research supporting the importance of quality evaluation in terms of usability, security and reliability (Fernandez et al., 2011). This literature suggests that in addition to treatment expectations, those who access the website have usability expectations. If these expectations are incongruent with the experience of

using the online platform the effect on engagement may be negative. Thus, it is important to understand the user experience so that we, as researchers can adequately describe what can be expected of the intervention in our participant information.

6.2.1 Aims

The primary aim of this trial was to evaluate the feasibility and preliminary efficacy of the online, *Feeling Better* pain management programme (McManus & McGuire, 2010) for pre-adolescent children with chronic pain and their care-givers.

6.2.2 Specific objectives

1. The primary objective of the present study was to evaluate the feasibility and piloting of the waitlist controlled, *Feeling Better* trial in terms of recruitment, adherence (engagement with the intervention), attrition from therapy and drop out (i.e. loss to follow-up as measured by missing post-intervention data), acceptability and treatment satisfaction.

2. A secondary objective was to assess the potential effectiveness of the *Feeling Better* programme to inform a future RCT. This was achieved by examining changes in clinical outcomes from baseline, among those in the internet intervention group compared to those in the control group at post-intervention and three-month follow-up. The following clinical outcomes were evaluated:

- Physical functioning (limitations and health) - assessed using the PedsQL Quality of Life Inventory - Physical Health Subscale (primary outcome)
- Pain characteristics (intensity, duration and quality) – assessed using the Wong-Baker FACES Pain Rating Scale and numerical rating scales (primary outcome)
- Social and emotional functioning - assessed using the PedsQL Quality of Life Inventory - Psychosocial Health Subscale (PedsQL 4.0; Varni, 1998) (secondary outcome)

- Health related Quality of Life – assessed using the PedsQL Quality of Life Inventory (secondary outcome).
- Use of coping strategies for pain management – assessed using the PedsQL – Coping Skills Inventory (secondary outcome).

3. The final objective was to explore change in process variables that may contribute to potential therapeutic change in the above clinical outcomes. Evaluation was based on change in outcome measures between intervention and waitlist control groups at baseline, post- intervention and three-month follow-up. The following process variables were evaluated:

- Self-efficacy – assessed using the Self-efficacy for Functioning Despite Chronic Pain, Parent and Child version
- Pain catastrophising assessed using the Pain Catastrophising Scale – Parent and Child version)
- Parental protectiveness assessed using the Adult Response to Children’s Symptoms – Protect Subscale

6.2.3 Research questions

These objectives informed the following research questions:

1. What are the likely recruitment and retention rates across intervention groups, over 9 weeks, for a trial comparing remotely delivered, CBT-based, pain management support for pre-adolescent children with chronic pain and their care-givers compared to a usual care, waitlist control condition?
2. What are the individual patterns of website-use among those engaging with the website and those who fail to?
3. How acceptable is the *Feeling Better* website in terms of ease of use, understanding and relevance to children with chronic pain and their care-givers?
4. What are the changes from baseline, if any, in patient-centred

measures?

6.2.4 Hypotheses

First, it was hypothesized that the intervention would show evidence of feasibility (usability, acceptability and satisfaction) as demonstrated by high adherence, low attrition and participant satisfaction with the intervention upon completion. Second, it was hypothesised that children receiving the *Feeling Better* Internet intervention would show greater improvement in symptoms and modification of maladaptive pain cognitions compared to children in a waitlist control condition. It was hypothesised that these improvements would be maintained in children receiving the Internet intervention at the 3-month follow-up.

6.3 Methods

6.3.1 Ethical approval

As stated previously, this trial was approved by the Research Ethics Committee at National University of Ireland Galway [(Re. (13/ Nov/01)]. Written consent was obtained from parent participants and written assent was obtained from child participants who were willing to participate in accordance with the Declaration of Helsinki (Appendix 18 and 19). In line with the code of ethics governing this research, participants were reminded of the voluntary nature of the participation, about their right to withdraw at any time and that their data would be stored securely and anonymously in accordance with the Data Protection Act (1990).

6.3.2 Recruitment and uptake

6.3.3 Recruitment

The aim was to recruit approximately 70 children aged 5-12 years with non-malignant chronic pain and their care-giver(s) from the community, to be randomized to receive the intervention or a waitlist control condition (usual care). Recruitment took place from January 2016 – January 2017 and involved both traditional print methods i.e. press release and poster (Appendix 20 and 21) and online recruitment strategies.

Traditional recruitment methods included the use of national media and promotional materials to recruit from the community. Charities and parent support networks for families coping with chronic pain-related conditions were contacted and asked to promote the study. Researchers also recruited in person at organised events for children with chronic pain. Promotional materials including information letters, posters and flyers were mailed to schools, primary care clinics and private physiotherapy clinics.

Online recruitment methods were employed and conducted concurrent with traditional methods. These included the use of social media platforms such as Facebook, Instagram, Twitter and LinkedIn. From each social media platform, potential participants were directed to the host intervention website to learn more about the study or contact the research

team directly. Promotional materials were adapted for each social media platform to appeal to the target population i.e. children on Instagram or parents on Facebook. This was a cost-effective and accessible means of promoting the study. The host website supported this approach by providing printable participant information sheets, information about the research team, contact information and guided navigation through baseline screening and assessment procedures.

6.3.4 Eligibility

6.3.5 Inclusion/exclusion criteria

Study eligibility was assessed using the computerised questionnaires. Child participants were eligible if they: (1) were aged age 5 to 12 years, (2) experienced chronic or recurrent non-malignant pain present for a period of 3 months or more which met the criteria for a diagnosis of chronic or recurrent pain as defined by the International Association for the Study of Pain (IASP, 1986), (3) experienced pain at least once per week, (4) experienced pain interference in at least one area of daily functioning as per parent report (5) could read and write English, (6) had regular access to a computer with an Internet connection and (7) agreed not to engage in psychological treatment for chronic pain management during the active phase of participation. We chose the IASP definition because this is a globally recognised and internationally used definition.

6.3.6 Exclusion criteria

Participants were ineligible if: (1) the child had a serious psychiatric illness, (2) the child had pain associated with a chronic medical condition (e.g. cancer), (3) the child had a developmental disability which would prevent them understanding the research materials, (4) the parent or child was non-English speaking or (5) the family did not have regular access to the Internet on a computer or portable device e.g. tablet or laptop computer.

6.3.7 Study design

The was an exploratory, pilot randomised controlled trial to determine the feasibility, acceptability and potential efficacy of the online version of the Feeling Better intervention. This study uses a single-blinded, variable block, randomised parallel group design (ISRCTN58820406). Blinding of the participants and the research co-ordinator was not possible due to the nature of the intervention. Potential sources of bias were minimised where possible for example by collecting baseline data prior to randomisation and ensuring concealment by using an automated randomisation and group allocation process embedded within the online programme.

6.3.8 Randomisation

Assessments were completed online through a secure, password-protected website. Assessments were completed independently by children and parents at baseline before randomization, upon completion of the 9-week intervention and at a three-month follow-up period. Randomization was implemented using a computer-generated randomization schedule to derive a randomization assignment to the treatment conditions in blocks of 2, 4 and 6 for each ID number. The randomization assignment algorithm which produced group allocation was programmed into the *Feeling Better* system by a software engineer. This process was automatic and done immediately upon completion of the pre-treatment assessments, the group assignment was then notified to each participant on the website with instructions on how to proceed. At this point the research co-ordinator also received an email alert which identified group assignment.

6.3.9 Statistical analysis

Sample size estimates for this trial adhere to the rule of thumb proposed by Viechtbauer et al. (2015). An estimated sample of 59 participants would be required to investigate the feasibility and potential effectiveness of the *Feeling Better* intervention and to detect unforeseen problems such as ambiguities in inclusion criteria or misinterpretation of

questionnaire items. The proposed sample size is also consistent with much of the literature in this area and in line with the purpose of the aims of this trial. All results presented are based on intent-to-treat approach using the last observation carried forward method for imputation of missing values. All statistical analyses were performed using SPSS Statistics version 17.0 for Windows.

Demographic characteristics were summarised using descriptive statistics including means, SDs and frequencies. As recommended by the 2001 Consolidated Standards of Reporting Trials (CONSORT) statement, adjusted and unadjusted descriptive statistics are reported on all outcomes (Yu, Chan, Hopewell, Deeks, & Altman, 2010) Categorical variables were reported using frequency statistics. Continuous variables were reported using means and standard deviations. To verify the randomisation process and group equivalence, t-tests and chi square analyses were conducted. Hypothesis testing was carried out at the 5% level for primary and secondary outcomes. The two conditions (Internet intervention and waitlist control) were compared at pre-and post-intervention – using one-way between groups analysis of covariance (ANCOVA). This was intended to maximise the statistical power of the small sample and to check for differences in pre-intervention scores which could influence the outcome. Separate ANCOVAs were computed for the primary (pain and physical health) and secondary outcomes (psychological health, pain coping, quality of life, pain catastrophising, self-efficacy, parental protectiveness, treatment satisfaction). Group was entered as a fixed between-subject factor, and pre-intervention values were entered as a covariate. Preliminary checks were performed to ensure there was no violation of the assumptions associated with ANCOVA including normality, linearity, homogeneity of variances, homogeneity of regression slopes and reliable measurement of the covariate. Effect sizes for ANCOVAs are expressed as partial eta squared (η^2). These are interpreted using multivariate guidelines proposed by Cohen (1988) which outline a small effect size at 0.01, a medium effect size at 0.06, and as a large effect size when greater than or equal to 0.14 (pp. 284-7).

One-way ANOVA with repeated measures was used to explore the

maintenance of effects from T1 (prior to the intervention), through to T2 (following the intervention) and T3 (3-month follow-up) for the Internet intervention group. Separate one-way repeated measures ANOVAs were computed for all primary (pain and physical health) and secondary outcomes (psychological health, pain coping, quality of life, pain catastrophising, self-efficacy, parental protectiveness, treatment satisfaction).

6.3.10 Data Management

Questionnaire data was saved online using a LimeSurvey platform embedded within the intervention website. This data was automatically anonymised and stored separately i.e. in a separate database to the intervention content data (i.e. separate to user input into the website). All data was stored using industry standard protection and encryption procedures. Participant data was only accessible by the first author. When collated, this data was stored in encrypted virtual hard-drives, in both .csv and .sav file formats and made accessible to the trial supervisors (BMG, JE). This data will be retained for a period of 5 years in accordance with the NUI Galway data retention policy.

6.3.11 Participants and setting

This trial was undertaken in an online environment with support from the National University of Ireland Galway School of Psychology and Centre for Pain Research [(Re. (13/ Nov/01)].

Figure. 6.1 illustrates the process of enrolment of participants and its progression throughout the treatment study. As shown, 80 child-parent dyads expressed an interest in the study and were assessed for eligibility. A total of 3 dyads were excluded because they did not meet inclusion criteria ($n = 2$ due to ineligible age (>12 years), $n = 1$ due to ineligible condition). A further 10 families who initially expressed an interest in the study could not be reached (passive refusal) and did not complete pre-treatment assessments. The overall participation rate was 84%. A total of 67

participants were randomly assigned to the treatment condition, Internet intervention (n = 35) or wait-list control (n = 32). Usual medical care continued for both groups. Nineteen child-parent dyads (n = 9 Internet intervention group, n = 10 wait-list control group) did not complete post-treatment assessments. Three dyads in the Internet intervention and 1 dyad in the control condition actively withdrew early in the intervention phase (9-week treatment phase). Three dyads actively withdrew from the study (n = 2 Internet intervention group, n = 1 waitlist control group) to begin face to face psychological therapy as part of their usual treatment. A fourth dyad actively withdrew due to Internet access issues (Internet intervention group). 15 parent-child dyads (n = 6 Internet treatment group, n = 9 wait-list control group) passively withdrew from the study, did not complete post-treatment assessments and could not be contacted to ascertain a reason for drop-out. Thus, the final sample (completing all three assessment phases) was n = 10 and the attrition rate was 28%. Intent-to-treat analysis was used, meaning the numbers of cases included in primary and secondary outcome analyses were n=35 for the Internet treatment group and n=33 for the wait-list control group.



CONSORT 2010 Flow Diagram

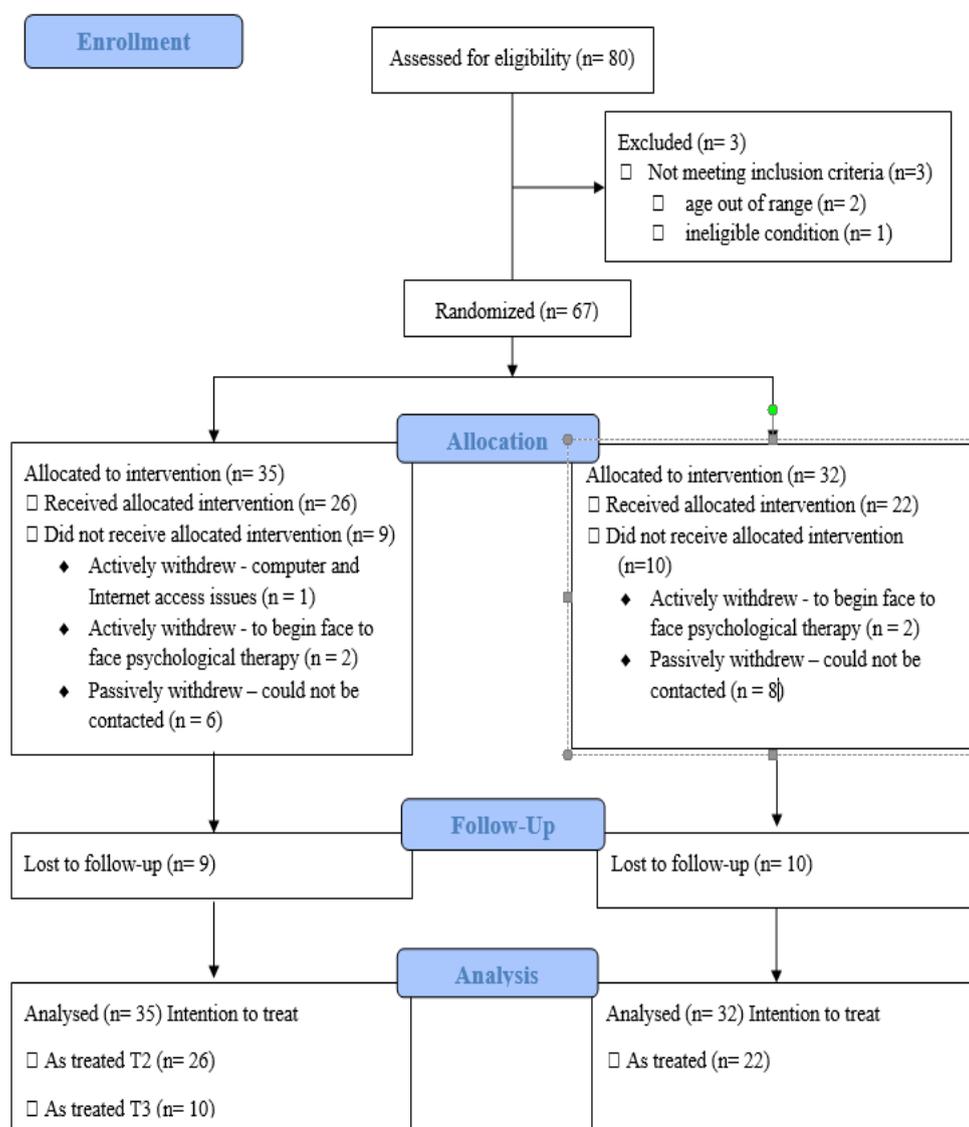


Figure 6.1. Flow of participants through the *Feeling Better* trial

6.3.12 Treatment condition

The *Feeling Better* programme was hosted on a dedicated, Amazon protected network. Participants' data and digital correspondence (email and instant messaging) were encrypted and separately and securely stored on the Amazon virtual web servers to guarantee privacy and confidentiality. The *Feeling Better* website-platform layout featured two structured pathways

though the programme. A structured therapy-based pathway specifically designed for pre- adolescent children and a separate information-based pathway which was designed for parents or care-givers. The *Feeling Better* programme was accessible using unique child and parent log-in codes. The therapeutic content of the programme was presented in the form of nine cognitive behavioural therapy-based interactive modules which were accessible in sequence and upon completion of the previous module (see Table 6.1). All participants received weekly support from an online tutor (the researcher) with previous experience of giving therapeutic support using remote delivery methods. Participants could log-in and compose and send e-mails and instant messages at any time. In line with the pre- defined treatment protocol, response to participant correspondence was individually tailored and submitted once per week and on set day per week. The only exception to this rule was correspondence relating to a technical or IT problem which was dealt with as quickly and efficiently as possible to avoid unnecessary interruption of treatment for the participant. The participant would also receive an immediate response to an emergency email. The telephone contact details for the principal investigator (BMG) and lead researcher (AT) were made available to all participants for the purposes of appropriate response to emergency situations.

Parents or care-givers had access to a parallel programme and received email correspondence with the same frequency as the children. The parents' platform consisted of the same module information content and correspondence options as that of the child platform. The content in the child platform was more interactive and personalized with automated tailored feedback, progress reports, multiple choice quizzes and video and audio material. Parents of all younger child participants (5 to 10 years) were instructed to coach their children, whereas parents of older child participants were encouraged to offer their assistance as required and encourage their children to take responsibility for their treatment. Increased use of coping/self-management skills was the aim of treatment and was presented as a primary of goal of the *Feeling Better* programme in the introductory section of the programme. A Certificate of Completion was issued to all

participants who completed the online programme (Appendix 22).

6.3.13 Waitlist control group

The child and parent participants in the control group were contacted by email at the 5 week point of the intervention period to answer any questions and to set up the post-treatment assessment. After post-treatment assessment, these patients were invited to participate in the *Feeling Better* programme. Each participant was given a unique username and password to the *Feeling Better* programme. All participants agreed not to engage in psychological treatment for chronic pain management during the active phase of this intervention (T1-T3).

6.3.14 Theoretical framework and intervention content

As described in Chapter 2, 3 and 4 the theoretical frameworks used to guide this intervention were cognitive-behavioural and social cognitive theory. The core components of CBT incorporated in the *Feeling Better* intervention were: (i) education about chronic pain – assessment and management (Mission 1 and throughout), (ii) relaxation training - deep breathing, progressive muscle relaxation, guided visualisation and recognizing stress (Mission 2 and throughout), (iii) being active - exercise and activity pacing (Mission 3 and throughout), (iv) attention management – distraction and changing pain sensations (Mission 4 and 5), (vi) managing stress and negative emotions – cognitive skills and re-framing (Mission 6), (vii) problem solving and sleep hygiene (Mission 7), (viii) communication skills (Mission 8) and (viii) future planning (Mission 9). Parent modules were an information-based replication of the child modules featuring more detailed pain management education, instructions on how to facilitate their child's pain self-management practice and practical tips for practice.

Table 6.1

Final content of the Feeling Better website

Topic	Summary of content
Introduction	This section introduces users to purpose of <i>Feeling Better</i> programme, the overall goals, the online tutor, the confidential nature of this programme, the overall layout (Missions 1-9) and the pirate theme including the code of conduct and commitment to the programme.
Mission 1: Learn More-Feel Better (Understanding Pain) †	This module encourages users to recognise their symptoms (types & effects of pain) and introduce the factors that influence symptoms (e.g. thoughts, behaviours, emotions), introduces goal setting (SMART goals), reward-setting, engage with case-examples and implementation intentions (if-then plans), identify barriers and facilitators to pain management, re-quiz (session summary) and its potential benefits.
Mission 2: Relax More-Feel Better (Relaxation Training) †	This module begins with the weekly check-in (recap), encourages users to set a weekly goal and reward, and introduces users to relaxation skills (deep (belly) breathing, video and instructions), case examples and implementation intentions (if-then plans), signs of stress, to identify barriers and facilitators to relaxation, a re-cap quiz and homework (practice a 10-minute relaxation skill).
Mission 3: Move More-Feel Better (Exercise & Activity Pacing) †	This module begins with the weekly check-in (recap), encourages users to set a weekly goal and reward, and introduces users to exercise and activity pacing skills and to full-body relaxation skills (progressive muscle relaxation video and instructions), to identify suitable and alternative activities, to identify "fun" exercises and pacing activities, benefits of paced activity, fear avoidance, case examples and implementation intentions (if-then plans), signs of stress, to identify barriers and facilitators to relaxation, a re-quiz (session summary) and homework (create activity pacing plan).
Mission 4: Focus More-Feel Better†	This module begins with the weekly check-in (recap), encourages users to set a weekly goal and reward, and introduces users to guided attention management skills (look, listen remember) and to guided visualisation (mind movie relaxation and instructions), case examples and implementation intentions (if-then plans), re-quiz (session summary) and homework (attention management skill).

Mission 5: Discover More- Feel Better†	This module begins with the weekly check-in (recap), encourages users to set a weekly goal and reward, and introduces users to guided attention management skills (reinterpret the experience of pain) and to guided visualisation (pain re-focusing relaxation and instructions), case examples and implementation intentions (if-then plans), re-quiz (session summary) and homework (attention management skill).
Mission 6: Explore More - Feel Better (Thinking & Feeling) †	This module begins with the weekly check-in (recap), encourages users to set a weekly goal and reward, and introduces users to cognitive restricting skills (linking events, thoughts and feelings, identify evidence for and against helpful and unhelpful thoughts), identify coping skills, case examples and implementation intentions (if-then plans), list coping skills statements, re-quiz (session summary) and homework (cognitive self-monitoring exercise).
Mission 7: Solve More - Feel Better (Problem solving) †	This module begins with the weekly check-in (recap), encourages users to set a weekly goal and reward, and introduces users to problem solving skills (problem solving plan, troubleshooting), identify positive negative attitude, case examples and implementation intentions (if-then plans), family and sleep problem solving, re-cap guided visualisation skills, re-quiz (session summary) and homework (problem solving exercise).
Mission 8: Talk More - Feel Better) Communication†	This module begins with the weekly check-in (recap), encourages users to set a weekly goal and reward, and introduces users to positive communication skills (I sentence, BEST plans), brainstorming, communication skills with health professionals, case examples and implementation intentions (if-then plans), re-quiz (session summary) and homework (brainstorming exercise).
Mission 9: Practice More Feel Better (Future Planning) †	This module begins with the weekly check-in (recap), advice for staying on course, re-cap all skills "best bits" (core points) and identify barriers and facilitators to practice. Completers receive certificate of completion.
Compass	Homework section for easy access to weekly homework assignments
Pirate Pouch	Section containing printable pdfs to encourage further practice in the manner preferred by the user
Useful info and links	Information section containing links to local and international support networks and sources of information
Email reminders	Emails reminders sent every Friday to parents to encourage practice and give feedback on weekly progress. Internal emails (messages) sent to child participant every Friday to encourage progress.

†All users are directed through these core modules in sequence (tunnelled navigation).

6.3.15 Outcome Measures

Feasibility and clinical outcome assessments were carried out at baseline (T1), at post-treatment (T2, 9-11 weeks) and at three-month follow-up (T3) (see Appendix 23 for copy of full assessment questionnaires). The feasibility outcomes were recruitment, adherence and participant evaluations. The clinical outcomes were physical functioning, pain intensity, emotional distress (anxiety and depression) and quality of life.

6.3.16 Primary outcome measures – feasibility measures

The primary feasibility outcome measures for this study are: recruitment and retention rates, treatment engagement, website satisfaction, treatment expectancy, treatment acceptability and satisfaction.

6.3.17 Recruitment and retention

Recruitment was measured by the percentage of eligible participants who progressed from initial enquiry to treatment allocation and enrolment. Retention to follow-up was measured by the completeness of online data collection in each arm at post-intervention and 3- month follow-up. This will be presented as a percentage of the participants randomised.

6.3.18 Adherence and website use (engagement)

Treatment compliance was measured at 9-11 weeks post-randomisation by means of objective system usage data ('participant access data'). In line with similarly designed interventions (Palermo et al., 2009; Bos et al., 2013), website use and treatment engagement were measured in the following ways:

- the number of sessions completed (0-18 total). treatment compliance)
- the number of times users logged in ('unique logins')
- the average length of time users remained logged in
- the number of times individual pages were viewed

- the number of interactive strategies completed
- the number of participants who submitted weekly homework
- the most and least accessed website components based on the module page accessed and completion rates

Evaluation also emulated another online trial whereby on a set day, each week (Monday), module completion rates were evaluated and counts of complete answers were expressed as a weekly percentage (Moss-Morris et al., 2012). Completers were deemed those who completed five or more modules. However, unlike similar trials, it was decided that while the submission of homework would be encouraged it would not be necessary to progress to the next module.

6.3.19 Treatment satisfaction and acceptability

Treatment satisfaction and acceptability was measured using the Internet Evaluation and Utility Questionnaire (Thorndike et al., 2008). This 15-item measure of user experience and perceptions of the internet intervention was selected because it is closely aligned to the Model for Internet Interventions (Ritterband et al., 2009) which guided intervention development. Earlier, short-form versions of this measure have shown good internal reliability ($\alpha=0.69$) (Thorndike et al., 2008). Participants are asked to rate their user experience on a 5- point Likert scale ranging from 0 (not at all) to 4 (very), with two open-ended items requesting patients to identify ‘most helpful’ and ‘least helpful’ parts of the website.

6.3.20 Website satisfaction

Website satisfaction was further measured using six purposely constructed questions based on Ritterband’s model for intervention development (Ritterband et al., 2009). These consisted of open-ended, website specific preference questions referring to theme, navigation, intensity, interactive features, ease of use and level of tutor contact.

Participants are asked to rate their user experience satisfaction on a 5- point Likert scale ranging from 0 (not at all) to 4 (very).

6.3.21 Treatment expectancies

Post-randomisation, children and parents completed a measure of treatment expectancies which comprised of a visual analogue scale with anchors (0 = No help, 10 = Most help you can imagine) that asked participants to select the face and numerical anchor which shows how much they anticipate this programme might help with symptom improvement. Higher scores indicated more positive treatment expectancies.

6.3.22 Feasibility criteria and programme use outcomes

In line with Step 6 of the IM protocol guiding this research, the programme use outcomes for this study are: (i) parents adopt the *Feeling Better* intervention and respond favourably to participant recruitment procedures and (ii) families implement the *Feeling Better* intervention and facilitate regular use within the home (Table 6.2). This attempt to anticipate barriers to implementation and beneficial effect was informed by the preceding needs assessment.

Table 6.2

Feasibility criteria and programme use outcomes for adoption and implementation

Adoption use outcome: parents adopt the <i>Feeling Better</i> intervention and respond favourably to participant recruitment procedures	
Performance objective 1	Parents agree to participate and agree to allow their child to participate in the <i>Feeling Better</i> feasibility trial <ul style="list-style-type: none"> • Recruit target sample size • Demonstrate willingness to be randomised • Low pre-enrolment attrition rate • Moderate enquiry to registration conversion rate
Implementation use outcome: Families implement the <i>Feeling Better</i> intervention and facilitate use	
Performance objective 2	Online tutor(s) complete training in the content and delivery of the <i>Feeling Better</i> intervention
Performance objective 3	Parents and children agree to participate in the <i>Feeling Better</i> feasibility trial <ul style="list-style-type: none"> • Demonstrate willingness to be randomised • Achieve at least 70% completion rate • Report medium to high treatment satisfaction
Performance objective 4	Families prepare the home environment to facilitate use of the Internet and computer and the practice of CBT skills with some level of quiet or privacy <ul style="list-style-type: none"> • Achieve small effect sizes
Performance objective 5	Local agencies, charities and/or clinics support participant recruitment to the <i>Feeling Better</i> intervention within the feasibility trial <ul style="list-style-type: none"> • Achieve at least a moderate conversion rate • Achieve a low pre-enrolment attrition

6.3.23 Primary outcome measures – clinical measures

6.3.24 Physical health (limitations)

Physical health (functional limitations) were assessed using the Pediatric Quality of Life Inventory - Physical health subscale (PedsQL™ 4.0; Varni, 1998). The physical health summary scale produced a summary

score for child physical functioning based on 8 items assessed on a five-point (ages 8-12 years and adult report) and three-point (ages 5-7 years) Likert scale.

6.3.25 Pain intensity

Pain intensity was assessed using the Wong-Baker FACES Pain Rating Scale, a six-item ordinal faces scale (WBS) which assessed the intensity of pain from none to worst in the previous two weeks. Previous research exploring the reliability of this measure reported good internal consistency coefficients for this scale on the child-report version, ranging from $\alpha = 0.83$ for children ages 3-7 to $\alpha = 0.96$ for children ages 8-12 (Keck, Gerkenmeyer, Joyce & Schade, 1996).

6.3.26 Secondary outcome measures – clinical measures

6.3.27 Mood

Mood was assessed using the Pediatric Quality of Life Inventory - Psychological Health Subscale (PedsQL™ 4.0; Varni, 1998). The psychological health summary scale produced a summary score for child emotional, social and role functioning based on 15 items assessed on a five-point (ages 8-12 years and adult report) and three-point (ages 5-7 years) Likert scale.

6.3.28 Health -related quality of life

Child health-related quality of life was assessed using the Pediatric Quality of Life Inventory -(PedsQL™ 4.0; Varni, 1998). The PedsQL™ 4.0 generic core scale produced a total summary score for child health related quality of life based on 23 items assessed on a five-point (ages 8-12 years and adult report) and three-point (ages 5-7 years) Likert scale. Previous research exploring the reliability of this measure reported the internal consistency coefficients for the three overall scores on the child-report

version as ranging from 0.80 to 0.89 (Varni et al., 2001; Varni, Burwinkle, Katz, Meeske, & Dickinson, 2002).

6.3.29 Pain coping skills

Child use of strategies to cope with pain was measured using the Pediatric Quality of Life-Pediatric Pain Coping Inventory (PedsQL-PCI; Varni, 1996). Previous research exploring the reliability of this measure reported good internal consistency coefficients for the overall scale ($\alpha = 0.85$) (Varni et al., 1996).

6.3.30 Pain catastrophizing

Level of catastrophizing was assessed with the Pain Catastrophising Scale-Child and Parent versions (PCS-C&P; Crombez et al., 2003). The parent and child version of the PCS each contain 13 items on which participants are asked to report using a 5-point rating scale. The PCS-C has shown good internal consistency ($\alpha = 0.88$) with adequate predictive validity (Crombez et al., 2003; Morris, Nagel, Heinrich, & Kroëner-Herwig, 2006).

6.3.31 Self-efficacy for functioning despite pain

Pain related self-efficacy was assessed using the Self-efficacy for Functioning Despite Pain Scale – Child and Parent report (Bursch et al., 2006). The scale has shown good internal consistency for child ($\alpha = 0.89$) and parent ($\alpha = 0.90$) versions and strong evidence for construct validity in 23 of 27 hypothesised correlations (Bursch et al., 2006).

6.3.32 Parental protectiveness

Levels of parental protective behaviour was assessed using the Adult Response to Children's Symptoms–Protect Subscale-child and parent versions. The scale has shown good internal consistency for child ($\alpha = 0.86$)

and parent ($\alpha = 0.84$) versions (Walker, Ley & Whitehead, 2006).

6.3.33 Demographic variables

Socio-demographic characteristics and condition-specific background information for the participants in each arm of the trial were recorded at baseline to determine if there were any inequalities across groups in terms of uptake, randomisation, engagement and retention. This data included age, sex, ethnic group, marital status, highest level of educational attainment, pain type and duration.

6.3.34 Adverse events

Participants were asked to respond to an open-ended question concerning potential adverse events occurring during the study at post-intervention and follow-up assessment.

6.4 Results

6.4.1 Sample Characteristics

The demographic and pain-related characteristics of the sample are presented in Table 6.3 and 6.4. Participants were 67 children (23 males and 44 females) aged between 5 and 12 years ($M = 9.1$, $SD = 1.97$) and 67 caregivers (57 mothers, 9 fathers and 1 grandmother), predominantly female (9 males and 58 females) and aged between 30 and 50 years ($M = 40.0$, $SD = 4.90$) (see Tables 6.3-6.4).

Table 6.3
Child and parent demographic characteristics at baseline (before randomization).

Child demographic characteristics	Internet treatment (n = 35)	Waitlist control (n= 32)	Total (n = 67)
Sex (n (%) female)	26 (74.3)	18(56.3)	44 (65.7)
Age (mean, SD)	8.8 (2.04)	9.5 (1.85)	9.1 (1.97)
5–7 years: n (%)	8 (22.9)	7 (21.9)	15 (22.4)
8–12 years: n (%)	27 (77.1)	25 (78.1)	52 (77.6)
Primary pain problem: n (%)			
Head	9 (25.7)	6 (18.8)	15 (22.4)
Abdomen	5 (14.3)	5 (15.6)	10 (14.9)
Musculoskeletal	10 (28.6)	14 (43.8)	24 (35.8)
Multiple	11 (31.4)	7 (21.9)	18 (26.9)
Primary pain location: n (%)			
Head	16 (45.7)	18 (56.3)	34 (50.7)
Abdomen	10 (28.6)	9 (28.1)	19 (28.4)
Chest	2 (5.7)	4 (12.5)	6 (9)
Back	11 (31.4)	10 (31.3)	21 (31.3)
Shoulder	8 (22.9)	4 (12.5)	12 (17.9)
Hips	10 (28.6)	6 (18.8)	16 (23.9)
Neck	10 (28.6)	9 (28.1)	19 (28.4)
Arm	7 (20)	6 (18.8)	13 (19.4)
Hands	8 (22.9)	7 (21.9)	15 (22.4)
Legs	12 (34.3)	10 (31.3)	22 (32.8)
Knees	9 (25.7)	9 (28.1)	18 (26.9)
Ankles	7 (20)	5 (15.6)	12 (17.9)
Feet	8 (22.9)	5 (15.6)	13 (19.4)
Other Pain	8 (22.9)	7 (21.9)	15 (22.4)
Location			
Pain frequency: n (%)			
1–2 /week	4 (11.4)	5 (15.6)	9 (13.4)
3–6 /week	8 (22.9)	9 (28.1)	17 (25.4)
Daily	23 (65.7)	18 (56.3)	41 (61.2)

Table 6.4
Parent demographic characteristics at baseline (before randomization).

Parent demographic characteristics	Internet treatment (n = 35)	Waitlist control (n= 32)	Total (n = 67)
Sex (n (%) female)	30 (85.7)	28 (87.5)	57 (85.1)
Age mean (SD)	39.6 (4.60)	40.6 (5.24)	40.0 (4.90)
Nationality, %			
Irish	29 (82.9)	27 (84.4)	56 (83.6)
British	1 (2.9)	0	1 (1.5)
American	3 (8.6)	4 (12.5)	7 (10.4)
Canadian	1 (2.9)	0	1 (1.5)
Other	1 (2.9)	1 (3.1)	2 (3)
Missing	0	0	0
Relationship status			
Single	3 (8.6)	1 (3.1)	4 (6)
Married	19 (54.3)	19 (59.4)	38 (56.7)
Co-habiting	6 (17.1)	3 (9.4)	9 (13.4)
Separated	6 (17.1)	8 (25)	14 (20.9)
Widowed	0	1 (3.1)	1 (1.5)
Divorced	1 (2.9)	0	1 (1.5)
Education, %			
Second level	12 (34.3)	6 (18.8)	18 (26.9)
Post second level	9 (25.7)	10 (31.3)	19 (28.4)
Third level	14 (40)	14 (50)	30 (44.8)
Missing	0	0	0
Employment status, %			
Full time	12 (34.3)	11 (34.4)	23 (34.3)
Part time	5 (14.3)	12 (37.5)	17 (25.4)
Homemaker	14 (40)	7 (21.9)	21 (31.3)
Student	1 (2.9)	2 (6.3)	3 (4.5)
Unpaid	1 (2.9)	0	1 (1.5)
Unemployed	2 (5.7)	0	2 (3)
Other	0	0	0

The Internet and control groups were equivalent with respect to age, sex, pain condition, parent marital status and parent education. Independent samples t tests indicated that participants in the Internet intervention and control group were also equivalent on pre- intervention measures of pain intensity, physical health, psychosocial health, quality of life, pain

catastrophising, perceived self-efficacy for functioning despite pain and parental protectiveness.

6.4.2 Study Completers and Drop-outs

An independent samples t-test demonstrated statistically significant differences in baseline report of clinical outcomes among those children and parents who completed the study and those who were lost to follow-up, across intervention conditions (intervention and control) and within the Internet intervention group (see Table 6.5 and 6.6).

According to child report, baseline levels of psychological health and overall quality of life were significantly lower and inferior among those children who completed the study compared to those who were lost to follow-up. Baseline levels of pain catastrophising were significantly higher and inferior among those who completed the study compared to those who were lost to follow-up. Baseline levels of sleep quality were significantly higher and indicative of lesser sleep quality among those who completed the study compared to those who were lost to follow-up.

According to child report, for those children assigned to the internet intervention, baseline levels of physical health (physical limitations) and psychological health were significantly lower and inferior among those children who completed the *Feeling Better* programme compared to those who dropped out of the Internet intervention. Baseline levels of sleep quality were significantly higher and inferior among those who completed the *Feeling Better* programme compared to those who dropped out of the Internet intervention.

According to parent report, baseline levels of psychological health, pain catastrophizing and sleep quality were significantly inferior among those children who completed the study compared to those who were lost to follow-up.

According to parent report, for those children assigned to the Internet intervention, baseline levels of sleep quality were significantly higher and inferior among those who completed the *Feeling Better* programme compared to those who dropped out of the Internet intervention.

6.4.3 Treatment expectancies

There were no significant differences in pre-treatment expectation of symptom improvement among children ($M = 5.07$, $SD = 2.59$) and parents ($M = 5.34$, $SD = 1.82$). An independent samples t-test also indicated there was no significant difference in treatment expectations between those in the Internet intervention group and the Waitlist control group as reported by children, $t(65) = .036$, $p = .97$, and parents, $t(65) = 2.65$, $p = .79$. There was also no significant difference in treatment expectations between children assigned to Internet group, who completed the programme ($n=26$) and those who failed to complete the programme (i.e. drop-outs; $n=9$), $t(33) = 0.31$, $p = .76$. According to parent report, there was a significant difference in treatment expectations among parents assigned to the Internet group, $t(33) = 2.63$, $p = .01$. Treatment expectations were significantly higher among those who dropped out of the Internet intervention group ($M = 6.67$, $SD = 1.94$) compared to those who completed the *Feeling Better* programme ($M = 4.96$, $SD = 1.59$).

Table 6.5

Baseline comparison of clinical outcomes among children who completed the study and those who were lost to follow-up.

Children	Completers (n=48)	Drop-outs (n=19)	<i>t</i>	<i>df</i>	<i>p</i>	Internet Intervention Completers (n = 26)	Internet Intervention Drop-outs (n = 9)	<i>t</i>	<i>df</i>	<i>p</i>
	Mean (SD)	Mean (SD)				Mean (SD)	Mean (SD)			
Physical health	25.78 (14.86)	34.21 (20.74)	1.86	65	.07	27.17 (15.63)	38.54 (19.01)	1.781	33	.08
Pain intensity	6.97 (1.98)	6.37 (2.28)	-1.07	65	.29	6.94 (1.77)	6.44 (2.21)	-0.68	33	.50
Psychological health	35.24(21.06)	47.28 (19.70)	2.15	65	.04	35.45 (23.58)	53.15 (18.68)	2.035	33	.05
Quality of Life	31.95 (15.94)	42.73 (18.99)	2.36	65	.02	32.57 (16.98)	48.07 (18.13)	2.321	33	.03
Pain coping	1.14 (0.31)	0.99 (.31)	-1.77	65	.08	1.13 (0.32)	0.92 (0.32)	-1.74	33	.09
Self-efficacy	26.38 (3.56)	25.00 (3.92)	-1.39	65	.17	26.19 (3.84)	23.78 (3.99)	-1.61	33	.12
Pain catastrophising	40.27 (7.60)	33.26 (7.89)	-3.37	65	.001	39.42 (8.25)	33.22 (8.64)	-1.92	33	.06
Parental protectiveness	2.23 (.85)	2.21 (.73)	0.10	65	.92	2.28 (0.80)	2.00 (0.81)	-0.90	33	.37
Sleep	7.83 (2.64)	6.00 (3.59)	-2.02	65	.05	7.69 (2.81)	4.44 (3.43)	-2.82	33	.01

Table 6.6

Baseline comparison of clinical outcomes among parents who completed the study and those who were lost to follow-up.

Parents	Completers (n=48)	Drop-outs (n=19)				Internet	Internet			
			<i>t</i>	<i>df</i>	<i>p</i>	Intervention Completers (n = 26)	Intervention Drop-outs (n = 9)	<i>t</i>	<i>df</i>	<i>p</i>
	Mean (SD)	Mean (SD)	<i>t</i>	<i>df</i>	<i>p</i>	Mean (SD)	Mean (SD)	<i>t</i>	<i>df</i>	<i>p</i>
Physical health	30.47 (15.29)	32.07 (22.31)	0.34	65	.74	30.89 (14.40)	36.11 (20.68)	0.84	33	.41
Pain intensity	6.00 (1.83)	5.29 (2.22)	-1.35	65	.18	5.92 (1.83)	4.89 (1.67)	-1.49	33	.15
Psychological health	36.08 (15.90)	46.84 (18.27)	2.40	65	.02	37.95 (17.55)	51.48 (19.45)	1.94	33	.06
Quality of life	34.13 (13.71)	41.70 (18.01)	1.86	65	.07	35.49 (14.83)	46.13 (18.74)	1.73	33	.09
Pain coping	1.13 (0.35)	0.95 (0.29)	-2.01	65	.05	1.13 (0.35)	0.95 (0.27)	-1.38	33	.18
Self-efficacy	25.92 (5.66)	24.53 (5.08)	-0.93	65	.36	24.69 (6.05)	23.89 (4.17)	-0.37	33	.72
Pain catastrophising	38.92 (10.73)	31.26 (12.99)	-2.48	65	.02	36.85 (11.97)	28.11 (11.84)	-1.89	33	.07
Parental protectiveness	2.23 (0.85)	1.86 (0.45)	-1.59	65	.12	2.14 (0.89)	1.94 (0.89)	-0.58	33	.57
Sleep	7.21 (2.32)	5.05 (2.78)	-3.24	65	.002	6.69 (2.46)	4.22 (1.86)	-2.74	33	.01

6.4.4 Primary feasibility outcome analyses: pre- to post-treatment results

6.4.5 Attrition

A total of 67 children and parents were assessed at baseline (Internet Intervention $n = 35$; Control condition $n = 33$), 48 dyads provided follow-up data at Time 2, and 10 dyads provided follow-up data at Time 3. In this study, the first block of attrition was 16% and comprised of those who enquired via email expressed initial interest but did not complete baseline measures ($n = 13$, “non-responders”). The second block of attrition was 28% and occurred between baseline and post-intervention follow-up – that is after randomisation to treatment group ($n=19$, “drop-outs”). The third and largest block of attrition was 71% and occurred at 3-month follow-up (“non-completers”). A total of nineteen dyads (Internet intervention, $n = 9$; Waitlist control, $n = 10$) were lost to follow-up at Time 2 and 16 dyads (Internet Intervention) were lost to follow-up at Time 3. Active withdrawal from the Internet Intervention group was due to computer and Internet access issues ($n = 1$ dyad) and initiation of face to face therapy elsewhere during the treatment phase of the intervention (Internet intervention, $n = 2$; waitlist control, $n = 1$). The remaining withdrawals (Internet Intervention $n=6$, Waitlist Control $n = 9$) were passive and participants could not be contacted to ascertain a reason. Passive withdrawal from the Internet Intervention group ($n=6$) may have been due to participants’ discomfort at not having engaged with the programme. Of the six dyads, four accessed the website only once.

A logistic regression analysis was performed to assess the impact of several factors on the likelihood of intervention loss to follow-up among child participants (Table 6.7). The dichotomous, dependent variable in this analysis was loss to follow-up (0= No, 1 = Yes). The predictor variables were group (intervention/control), sex (male/female), age, pain location (head, abdomen, musculoskeletal or multiple), pain intensity, pain coping, pain catastrophising, self- efficacy, parental protectiveness, physical health, psychological health, overall quality of life, sleep quality and treatment expectations. The full model containing all predictors was statistically significant $\chi^2 (15, N= 67) = 37.26, p < .01$ ($p = .001$) indicating the model

reliably predicted those who were lost to follow-up. The model explained between 42.7% (Cox and Snell R) and 61.2% (Nagelkerke R squared) of the variance and correctly classified 89.6% of participants. As shown in Table 6.5, four of the independent variables made a unique statistically significant contribution to the model (headache pain, parental protective behaviour, pain coping and physical health). The strongest predictor of loss to follow-up was over-protective parenting behaviour (Wald = 8.12, $p < .01$, OR = 129.12) suggesting participants whose parents reported high levels of parental protective behaviour were more likely to drop-out of the study.

Table 6.7
Logistic regression predicting likelihood of loss to follow-up

	β	SE	Wald	df	<i>p</i>	Odds Ratio	95% CI for Odds Ratio	
							Lower	Upper
Group	-1.6	1.06	2.27	1	.13	0.203	0.025	1.616
Sex	-1.26	1.02	1.52	1	.22	0.284	0.038	2.101
Age	0.261	0.26	1.05	1	.31	1.298	0.787	2.14
Pain Location			7.67	3	.05			
Headache	-3.95	1.5	6.91	1	.01	0.019	0.001	0.366
Abdomen	-0.6	1.56	0.15	1	.70	0.548	0.026	11.721
Musculoskeletal	-0.94	1.21	0.61	1	.44	0.391	0.037	4.165
Multiple								
Pain intensity	0.097	0.28	0.12	1	.73	1.102	0.637	1.908
Protective behaviour	4.861	1.71	8.12	1	.004	129.1	4.561	3655.5
Pain catastrophising	-0.11	0.08	2.18	1	.14	0.895	0.773	1.037
Pain coping	-6.74	2.93	5.29	1	.02	0.001	0	0.369
Self-efficacy	-0.2	0.21	0.98	1	.32	0.815	0.544	1.221
Physical health	0.08	0.04	4.23	1	.04	1.083	1.004	1.169
Psychological health	0.064	0.04	2.26	1	.13	1.066	0.981	1.159
Quality of life	0.219	0.2	1.18	1	.28	1.245	0.838	1.847
Treat expectations	0.104	0.22	0.22	1	.64	1.11	0.717	1.717
Sleep	-2.67	6.33	0.18	1	.67	0.069	0.717	1.717

6.4.6 *Feeling Better* Intervention Use

In accordance with the Consolidated Standards of Reporting Trial (CONSORT) eHealth checklist (Eysenbach, 2013), we analysed the use of the *Feeling Better* programme among those allocated to the online programme.

6.4.7 Module completion

Tables 6.8 and 6.9 outline the pattern of website use among all child and parent participants allocated to the Internet intervention. Children in the Internet intervention group demonstrated higher engagement with the programme based on module completion compared to parents. Seventy four percent of children in the Internet intervention group ($n = 35$) completed 5 or more modules in the *Feeling Better* intervention. Children completed an average of 6.5 out of 9 modules ($SD = 3.24$) and parents completed an average of 3 out of 9 ($SD = 1.50$). Among those allocated to the Internet intervention group, a total of 19 children (54%) and none of the parents completed all 9 modules. Most of the child participants categorised as ‘completers’ were aged 9-11 years. Figure 6.2 illustrates the number of logins and modules completed by all child participants allocated to the Internet intervention group ($n=35$). Figure 6.3 illustrates the number of logins and modules completed by all parent participants allocated to the Internet intervention group ($n=35$). The usage pattern suggests that among children, those who logged in more than once tended to use the website overall for longer. Qualitative feedback collected at Time 2 suggests parents preferred to follow their child’s progress via the child pathway through the programme rather than access their own information-based, less interactive pathway.

Table 6.8

Website usage among those children in Internet intervention group (n=35)

	N	%	Mean (SD)	Range
Number (%) of eligible child participants who logged in	35	100		
Mean number of unique logins per child participant			13.86 (8.03)	1-24
Mean time spent logged in per module - mins			24.00	2-67
Number of child participants who accessed each module				
Introduction	35/35	100		
Mission 1	35/35	100		
Mission 2	31/35	89		
Mission 3	27/35	77		
Mission 4	26/35	74		
Mission 5	26/35	74		
Mission 6	22/35	63		
Mission 7	21/35	60		
Mission 8	20/35	57		
Mission 9	19/35	31		
Average number of modules completed by children			6.49 (3.18)	

Table 6.9

Website usage among those parents who completed the study (n = 35)

	N	%	Mean (SD)	Range
Number (%) of eligible child participants who logged in	35	100		
Mean number of unique logins per parent participant			3.46 (2.00)	1-7
Mean time spent logged in per module - mins (range)			6.00	2-15
Number of parent participants who accessed each module				
Introduction	35/35	100		
Mission 1	35/35	100		
Mission 2	26/35	74		
Mission 3	22/35	63		
Mission 4	16/35	46		
Mission 5	7/35	20		
Mission 6	0/35	0		
Mission 7	0/35	0		
Mission 8	0/35	0		
Mission 9	0/35	0		
Average number of modules completed by parents			3.03 (1.50)	

6.4.8 Pattern of website use

Among child ‘completers’ (n = 26), each module accessed was visited at least twice. Among parent ‘completers’ (n=7), each module accessed was visited at least once. The tunnelled navigational format of the website dictated progression by participants through the Introduction and 9 core modules. Optional sections included the Compass, Pirate Pouch, Pirate Post (message centre) and Instant Messaging application. Among children allocated to the Internet intervention group, the most accessed modules based on average number of times a given page was accessed were Exercise and Activity Pacing module (Mission 3), Pain Education module (Mission 1), the Attention Management 2 – changing pain sensations, (Mission 2), Attention Management 1– distraction, the Relaxation module (Mission 2), the Thoughts and Feelings module (Mission 6) (see Figure 6.2). The least popular modules among children were the Attention Management, Problem-solving and Future planning modules (see Figure 6.2). Parent usage data was limited. Of the modules completed, the most popular, based on number of visits were the Pain Education module (Mission 1), Relaxation module (Mission 2) and Exercise and Activity Pacing module (Mission 3). The least popular, least accessed modules were the Attention Management 1 and 2 modules. (see Figure 6.3). Interestingly almost all child participants took longer to complete the programme than the 9-week active intervention phase originally intended. Progression through the programme was slow but continuous.

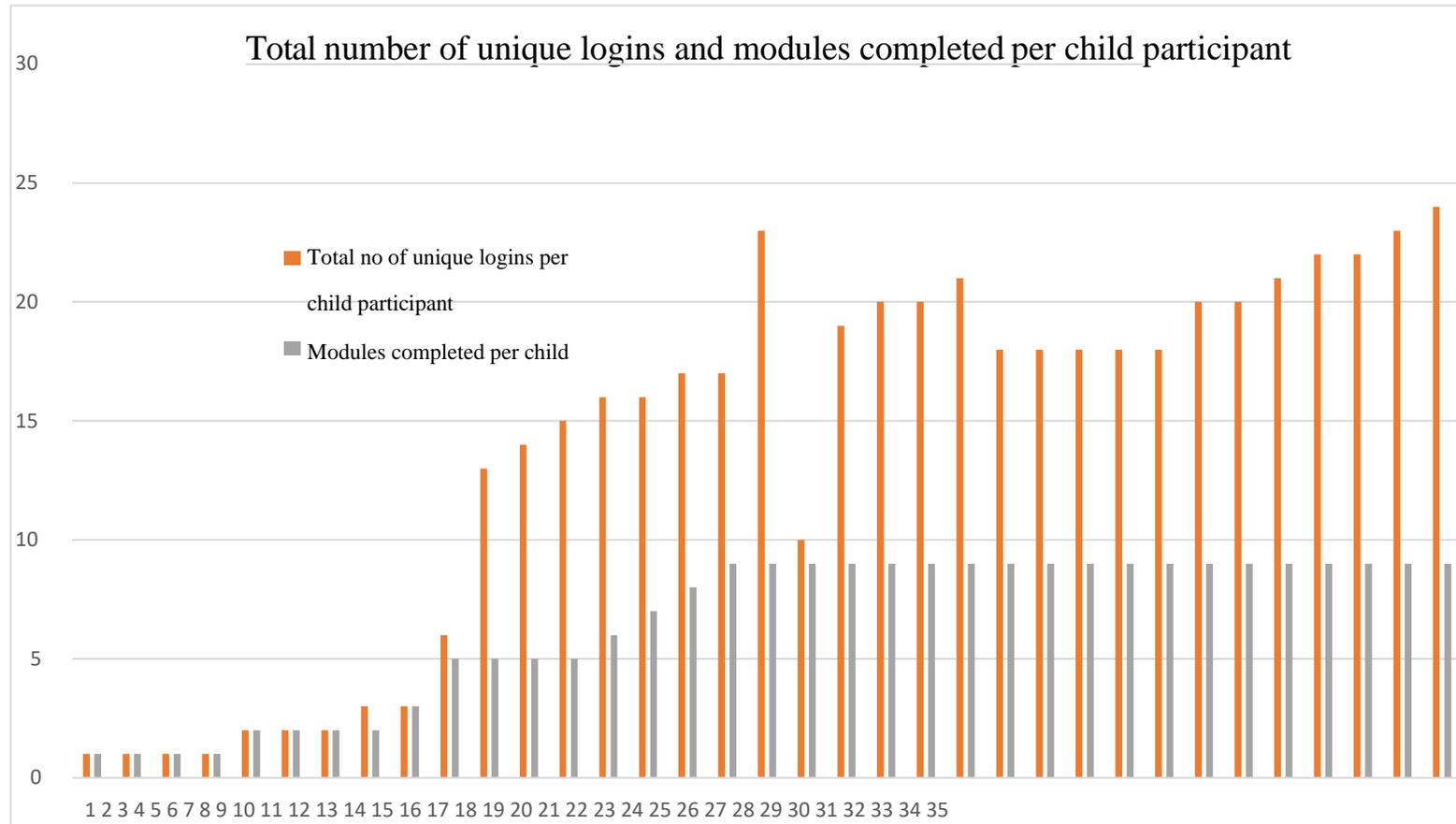


Figure 6.2 Total number of unique logins and modules competed per child participant.

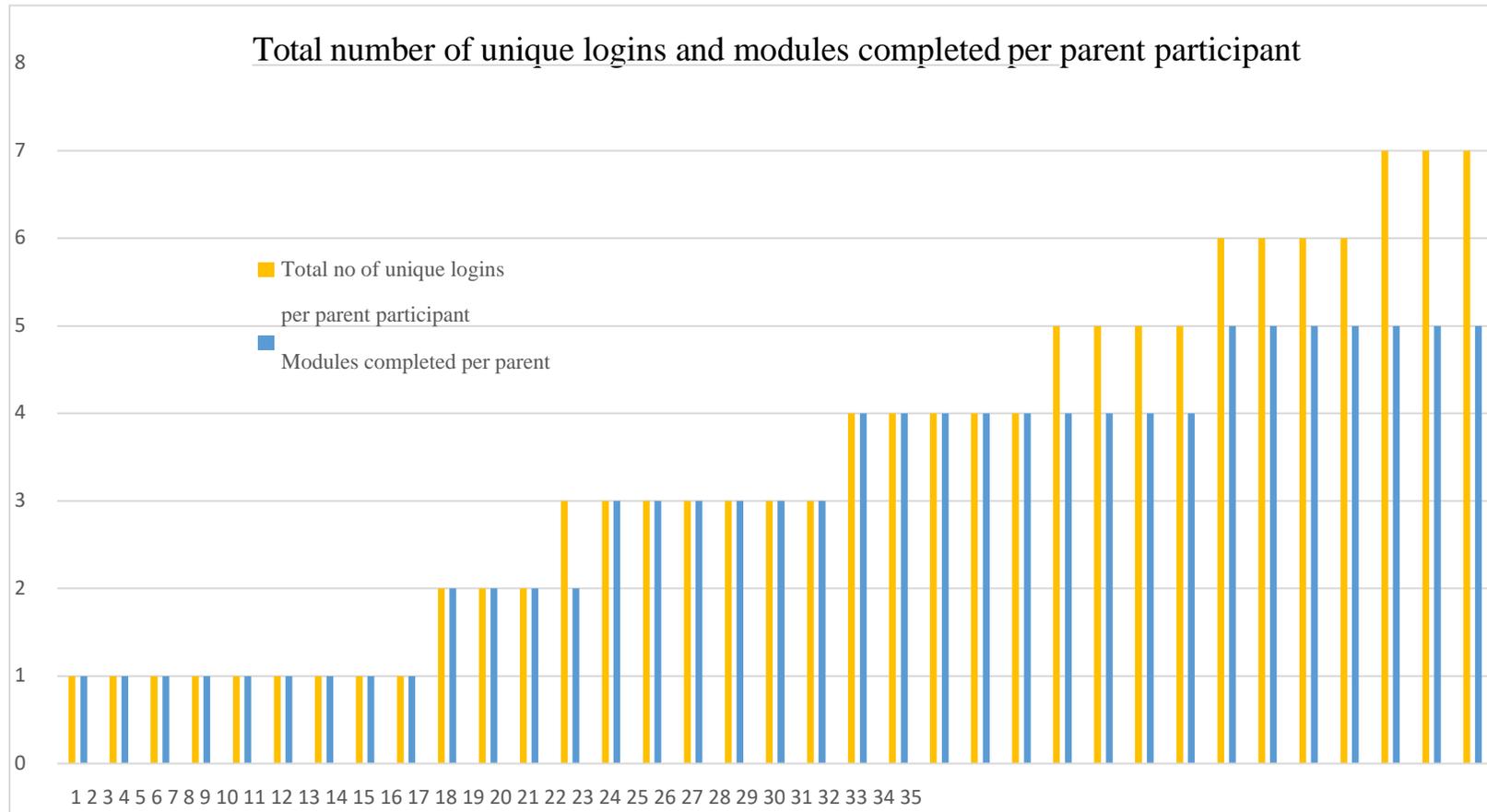


Figure 6.3 Total number of unique logins and modules completed per parent participant.

Within the core modules, the most accessed pages and components (embedded strategies or elements) of the programme, based on child participant access and completion rates were the ‘goal setting’ and ‘progress records’, the post-patient narratives, ‘Your Story’ exercises followed by the end of module ‘Quiz’ and the relaxation videos. The least popular components, based on completion, was the cognitive re-framing (Module 6) and problem- solving strategies (Module 7). Objectively collected usage data suggests the future planning session (Module 9) was popular but qualitative feedback in response to open-ended treatment satisfaction questions suggests the future planning ‘best bits’ exercises in this module were considered difficult to complete and were accessed more often than others for that reason. Qualitative feedback supports objectively collected usage data on all other modules and components.

6.4.9 Participant Feedback

Table 6.10 and Table 6.11 show participant evaluations of website acceptability, satisfaction and functionality. Children and parents rated the website as moderate to high in terms of usefulness, ease of use, relevance and appeal. Scores indicate the intervention and mode of delivery were considered acceptable, credible and relevant to children and parents. Participants response to open-ended questions on ease of use, theme, relevance and delivery support the trend in the objectively collected, website usability data which suggest those modules and components which were avoided, skimmed over or incomplete were considered most difficult to complete, less relevant or both. Except for the Future Planning module (Mission 9), qualitative feedback in response to the questions: *What was most helpful? What was least helpful?* suggest the most popular modules and intervention components were those which were most accessed and completed. Several suggestions for improvement were put forward. These include optional contact with peers and health professionals, further condensed and simplified content, more automated features and different modes of delivery for the parent pathway through the programme.

Table 6.10

Children's (n=26) ratings of overall website usability: N (%)

	Not at all	Slightly	Somewhat	Mostly	Very
	N (%)	N (%)	N (%)	N (%)	N (%)
How easy was the <i>Feeling Better</i> programme to use?				2 (7.7)	24 (92)
How (convenient) good was it to use the <i>Feeling Better</i> programme from home?			2 (7.7)	1 (3.8)	23 (88.5)
How much did the <i>Feeling Better</i> programme keep your interest and attention?			2 (7.7)	6 (23.1)	18 (69.2)
How much did you like the <i>Feeling Better</i> programme?			2 (7.7)	6 (23.1)	18 (69.2)
How much did you like the way the <i>Feeling Better</i> programme looked?			1 (3.8)	3 (11.5)	22 (84.6)
How worried were you about your privacy in using <i>Feeling Better</i> programme?	15 (57.7)	1 (3.8)	2 (7.7)	2 (7.7)	6 (23.1)
How (satisfied) happy were you with the <i>Feeling Better</i> programme?		1 (3.8)	1 (3.8)	5 (19.2)	19 (73.1)
How good of a fit was the <i>Feeling Better</i> programme for you?		1 (3.8)	1 (3.8)	3 (11.5)	24 (92)
How useful did you find the information in the <i>Feeling Better</i> programme?		1 (3.8)	1 (3.8)	4 (15.4)	20 (76.9)
How easy was the information to understand?			2 (7.7)	4 (15.4)	20 (76.9)
How much did you feel you could trust the information?	1 (3.8)	1 (3.8)		4 (15.4)	20 (76.9)
If your pain continues or returns, how likely would you be to come back to the <i>Feeling Better</i> programme?			2 (7.7)	3 (11.5)	21 (80.8)
How good of a method was the Internet for delivering this programme?			2 (7.7)	1 (3.8)	23 (88.5)

Table 6.11

Parents (n=26) ratings of overall website usability and evaluation data: N (%)

	Not at all	Slightly	Somewhat	Mostly	Very
	N (%)	N (%)	N (%)	N (%)	N (%)
How easy was the <i>Feeling Better</i> programme to use?				9 (34.6)	17 (65.4)
How convenient was <i>Feeling Better</i> programme to use?				10 (38.5)	16 (61.5)
How much did the <i>Feeling Better</i> programme keep your interest and attention?				13 (50)	13 (50)
How much did you like the <i>Feeling Better</i> programme?				13 (50)	13 (50)
How much did you like the way the <i>Feeling Better</i> programme looked?			2 (7.7)	11 (42.3)	13 (50)
How worried were you about your privacy in using <i>Feeling Better</i> programme?	11 (42.3)	5 (19.2)	1 (3.8)	6 (23.1)	3 (11.5)
How satisfied were you with the <i>Feeling Better</i> programme?			1 (3.8)	12 (46.2)	13 (50)
How good of a fit was the <i>Feeling Better</i> programme for you?			1 (3.8)	12 (46.2)	10 (38.5)
How useful did you find the information in the <i>Feeling Better</i> programme?			1 (3.8)	12 (46.2)	13 (50)
How easy was the information to understand?			1 (3.8)	13 (50)	12 (46.2)
How much did you feel you could trust the information?			3 (11.5)	11 (42.3)	12 (46.2)
If your pain continues or returns, how likely would you be to come back to the <i>Feeling Better</i> programme?		1 (3.8)	3 (11.5)	9 (34.6)	13 (50)
How good of a method was the Internet for delivering this programme?				11 (42.3)	15 (57.7)

6.4.10 Primary clinical outcome analyses: pre- to post-intervention results

Group differences in child and caregiver retrospective report of clinical outcomes were examined using baseline-adjusted ANCOVAs. Adjusted and unadjusted descriptive statistics for the primary and secondary outcome variables at each point of assessment are shown in Table 6.12 and Table 6.13.

6.4.11 Physical health

The baseline-adjusted ANCOVA used to evaluate group differences on child-reported mean physical health was significant, $F(1, 64) = 61.05$, $p < .001$, with a large effect size, partial $\eta^2 = .49$. Descriptive statistics suggest that mean physical health scores were higher and improved in the Internet intervention group ($M = 48.53$) relative to the waitlist control group ($M = 23.69$) (see Table 6.12).

The baseline-adjusted ANCOVA used to evaluate group differences on parent-reported, child mean physical health at post-intervention was also significant, $F(1, 64) = 41.81$, $p < .001$, with a large effect size, partial $\eta^2 = .40$. Mean post-intervention physical health values were larger in the Internet intervention group ($M = 48.10$) relative to the waitlist control group ($M = 24.93$) indicating greater improvement in parent-reported, child physical health among those in the Internet intervention group (see Table 6.13).

6.4.12 Pain intensity

The baseline-adjusted ANCOVA used to evaluate group differences on child-reported mean pain intensity at post-intervention was significant, $F(1, 64) = 31.97$, $p < .001$, with a large effect size, partial $\eta^2 = .33$. As shown in Table 6.12, the pattern of descriptive statistics was as expected, with the Internet intervention group having a smaller post-intervention mean ($M = 5.37$) relative to the waitlist control group ($M = 7.26$) indicating greater improvement in self-reported mean pain intensity among those in the

Internet intervention group.

The analysis of group differences on parent-reported, pain intensity at post-intervention was significant, $F(1, 64) = 35.80, p < .001$, with a large effect size, partial $\eta^2 = 0.36$. Mean post-intervention pain intensity values were smaller in the Internet intervention group ($M = 4.73$) relative to the waitlist control group ($M = 6.86$) indicating greater improvement in parent-reported, child mean pain intensity among those in the Internet intervention group (see Table 6.13).

6.4.13 Secondary clinical outcome analyses: pre- to post-treatment results

6.4.14 Psychosocial health

The baseline-adjusted ANCOVA used to evaluate group differences on child-reported psychosocial health at post-intervention was significant, $F(1, 64) = 70.41, p < .001$, with a large effect size, partial $\eta^2 = .52$. As shown in Table 6.12, descriptive statistics were as expected, with the Internet intervention group having a larger post-intervention mean ($M = 59.38$) relative to the waitlist control group ($M = 30.21$) indicating greater improvement in perceived psychosocial health among those in the Internet intervention group.

Similarly, group differences on parent-reported child psychosocial health at post-intervention was also significant, $F(1, 64) = 71.08, p < .001$, with a large effect size, partial $\eta^2 = 0.53$. Mean post-intervention psychosocial health values larger in the Internet intervention group ($M = 58.79$) relative to the waitlist control group ($M = 33.26$) indicating greater improvement in parent-reported child psychosocial health among those in the Internet intervention group (see Table 6.13).

6.4.15 Total quality of life

The baseline-adjusted ANCOVA used to evaluate group differences on child-reported total quality of life at post-intervention was significant, $F(1, 64) = 85.70, p < .001$ with a large effect size, partial $\eta^2 = .57$. As shown

in Table 6.12, mean post-intervention total quality of life values larger in the Internet intervention group ($M = 55.41$) relative to the waitlist control group ($M = 28.15$) indicating greater improvement in overall quality of life among those in the Internet intervention group.

The baseline-adjusted ANCOVA used to evaluate group differences on parent-reported quality of life at post-treatment were significant, $F(1, 64) = 71.90, p < .001$, with a medium effect size, partial $\eta^2 = .53$. Mean post-intervention quality of life values were larger in the Internet intervention group ($M = 54.97$) relative to the waitlist control group ($M = 30.47$) indicating greater improvement in parent-reported total quality of life among those in the Internet intervention group (see Table 6.13).

6.4.16 Pain coping

The baseline-adjusted ANCOVA used to evaluate group differences on child-reported pain coping at post-treatment was significant, $F(1, 64) = 5.43, p = .02$, with a medium effect size, partial $\eta^2 = .08$. As shown in Table 6.12, mean post-intervention pain coping values were as expected, with the Internet intervention group having a larger post-intervention mean ($M = 1.26$) relative to the waitlist control group ($M = 1.12$) indicating greater improvement in self-reported pain coping among those in the Internet intervention group.

The analysis of group differences on parent-reported, child pain coping at post-intervention were significant, $F(1, 64) = 7.80, p = .007$, with a medium effect size, partial $\eta^2 = 0.11$. Mean post-intervention pain coping larger in the Internet intervention group ($M = 1.26$) relative to the waitlist control group ($M = 1.11$) indicating greater improvement in parent-reported pain coping among those in the Internet intervention group (see Table 6.13).

6.4.17 Sleep quality

The baseline-adjusted ANCOVAs to evaluate group differences on child-reported sleep quality at post-treatment was significant, $F(1, 64) = 49.74, p < .001$, with a large effect size, partial $\eta^2 = 0.44$. As shown in Table

6.12, mean post-intervention sleep values were as expected, with the Internet intervention group having a smaller post-intervention mean ($M = 4.75$) relative to the waitlist control group ($M = 8.18$) indicating greater improvement in self-reported sleep among those in the Internet intervention group.

Similarly, group differences on parent-reported child sleep quality at post-intervention were significant, $F(1, 64) = 23.27, p < .001$, with a large effect, partial $\eta^2 = .27$. Mean post-intervention pain coping values smaller in the Internet intervention group ($M = 4.65$) relative to the waitlist control group ($M = 6.98$) indicating greater improvement in parent-reported child sleep quality among those in the Internet intervention group (see Table 6.13).

6.4.18 Self-efficacy

The baseline-adjusted ANCOVA used to evaluate group differences on child-reported self-efficacy for functioning despite pain at post-intervention was significant, $F(1, 64) = 80.87, p < .001$, with a large effect size, partial $\eta^2 = .56$. As shown in Table 6.12, mean post-treatment self-efficacy values were as expected, with the Internet treatment group having a smaller post-treatment mean ($M = 18.45$) relative to the waitlist control group ($M = 28.82$) indicating greater improvement in perceived self-efficacy for functioning despite pain among those in the Internet intervention group.

Similarly, group differences on parent-reported child-self-efficacy for functioning despite pain at post-intervention were significant, $F(1, 64) = 43.20, p < .001$, with a large effect size, partial $\eta^2 = 0.40$. Mean post-treatment self-efficacy values were smaller in the Internet treatment group ($M = 20.69$) relative to the waitlist control group ($M = 28.12$) indicating greater improvement in self-reported pain intensity among those in the Internet intervention group (see Table 6.13).

6.4.19 Pain catastrophising

The baseline-adjusted ANCOVA used to evaluate group differences

on child-reported pain catastrophising at post-intervention was significant, $F(1, 64) = 74.85, p < .001$ with a large effect size, partial $\eta^2 = .54$. As shown in Table 6.12, mean post-intervention pain catastrophising values were as expected, with the Internet treatment group having a smaller post-treatment mean ($M = 25.04$) relative to the waitlist control group ($M = 40.14$) indicating greater improvement in pain catastrophising among those in the Internet intervention group.

The baseline-adjusted ANCOVA used to evaluate group differences on parent-reported, child pain catastrophising at post-treatment were significant, $F(1, 64) = 56.71, p < .001$, with a large effect size, partial $\eta^2 = .47$. Mean post-treatment pain catastrophising values were smaller in the Internet treatment group ($M = 25.22$) relative to the waitlist control group ($M = 41.13$) indicating greater improvement in pain catastrophising among those in the Internet intervention group (see Table 6.13).

6.4.20 Parental protectiveness

The baseline-adjusted ANCOVA used to evaluate group differences on child-reported parental protective behaviour at post-treatment was significant, $F(1, 64) = 37.45, p < .001$, with a large effect size, partial $\eta^2 = .37$. As shown in Table 6.12, mean post-intervention parental protective behaviour values were as expected, with the Internet intervention group having a smaller post-treatment mean ($M = 1.67$) relative to the waitlist control group ($M = 2.47$) indicating greater improvement in child-proxy reports of parental protective behaviour among those in the Internet intervention group.

Similarly, group differences on parent-reported parental protective behaviour at post-treatment were significant, $F(1, 64) = 6.89, p = .011$, with a medium effect size, partial $\eta^2 = .10$. Mean post-intervention pain intensity values were smaller in the Internet treatment group having a smaller post-treatment mean ($M = 1.73$) relative to the waitlist control group ($M = 2.06$) indicating greater improvement in self-reported pain parental protective behaviour among those in the Internet intervention group (see Table 6.13).

Table 6.12.

Unadjusted and adjusted descriptive statistics on child-reported primary and secondary treatment outcomes

	Time	Intervention (n=35)					Control (n=32)						
		Mean	SD	Adjusted Mean	Std Error	95% CI		Mean	SD	Adjusted Mean	Std Error	95% CI	
						Lower	Upper					Lower	Upper
Pediatric Quality of Life Inventory - Physical Health Subscale													
Physical Health	Baseline	30.09	17.03					26.08	17.00				
	Post-treatment	49.64^{*#}	16.00	48.53^{*#}	2.19	44.15	52.90	22.46	16.43	23.69	2.29	19.11	28.26
	Follow-up	51.43^a	17.32					N/A	N/A				
Wong-Baker FACES Pain Rating Scale													
Pain Intensity	Baseline	6.81	1.87					6.78	2.29				
	Post-treatment	5.37[#]	1.56	5.37	0.23	4.90	5.83	7.25	1.65	7.26	0.24	6.77	7.74
	Follow-up	5.34	1.60					N/A	N/A				
Pediatric Quality of Life Inventory - Psychosocial Subscale													
Psychosocial Health	Baseline	40.00	23.51					37.19	18.73				
	Post-treatment	59.38[#]	12.99	58.97	2.33	54.32	63.62	30.21	17.19	30.66	2.44	25.79	35.52
	Follow-up	57.52	13.79					N/A	N/A				

SD: standard deviation; η^2 : partial eta squared; * denotes a significant change from baseline to post-treatment in a group, ^a denotes a significant change from baseline to 3-month follow-up in Internet group, # denotes a significant difference between groups at post-treatment.

Table 6.12.

Unadjusted and adjusted descriptive statistics on child- reported primary and secondary treatment outcomes (continued).

	Time	Intervention (n=35)					Control (n=32)						
		Mean	SD	Adjusted Mean	Std Error	95% CI		Mean	SD	Adjusted Mean	Std Error	95% CI	
						Lower	Upper					Lower	Upper
Pediatric Quality of Life Inventory - Overall Quality of Life													
Quality of Life Total Score	Baseline	36.55	18.35					33.32	16.45				
	Post-treatment	55.99 ^{*#}	11.89	55.41	2.03	51.36	59.47	27.51	15.26	28.15	2.12	23.91	32.39
	Follow-up	55.40 ^a	13.47					N/A	N/A				
Pediatric Pain Coping Inventory													
Pain Coping	Baseline	1.08	0.32					1.12	0.30				
	Post-treatment	1.25 ^{*#}	0.34	1.26	0.04	1.18	1.35	1.13	0.25	1.12	0.04	1.04	1.21
	Follow-up	1.24 ^a	0.32					N/A	N/A				
Adults' Response to Children's Symptoms (ARCS) – Protective Behaviour Subscale													
Parental Protectiveness	Baseline	2.21	0.80					2.25	0.84				
	Post-treatment	1.67 [#]	0.60	1.68	0.09	1.5	1.86	2.47	0.55	2.46	0.09	2.28	2.65
	Follow-up	1.65	0.60					n/a	n/a				

SD: standard deviation; η^2 : partial eta squared; * denotes a significant change from baseline to post-treatment in a group, ^a denotes a significant change from baseline to 3-month follow-up in Internet group, # denotes a significant difference between groups at post-treatment.

Table 6.12.

Unadjusted and adjusted descriptive statistics on child-reported primary and secondary treatment outcomes (continued).

	Time	Intervention (n=35)					Control (n=32)						
		Mean	SD	Adjusted Mean	Std Error	95% CI		Mean	SD	Adjusted Mean	Std Error	95% CI	
						Lower	Upper					Lower	Upper
Pain Catastrophizing Scale													
Pain Catastrophizing	Baseline	37.83	8.67					38.78	7.90				
	Post-treatment	24.91^{*#}	7.61	25.04	1.21	22.64	27.45	40.28	7.28	40.14	1.26	37.62	42.66
	Follow-up	25.74^a	8.49					n/a	n/a				
Self-efficacy for Functioning Despite Pain Scale													
Self-efficacy	Baseline	25.57	3.97					26.44	3.36				
	Post-treatment	18.23^{*#}	5.26	18.45	0.8	16.86	20.03	29.06	4.78	28.82	0.83	27.16	30.49
	Follow-up	18.69^a	5.08					n/a	n/a				
Numerical Rating Scale													
Sleep	Baseline	6.86	3.26					7.81	2.71				
	Post-treatment	4.57^{*#}	2.03	4.75	0.33	4.08	5.42	8.37	2.51	8.18	0.35	7.48	8.88
	Follow-up	4.46^a	2.38					n/a	n/a				

SD: standard deviation; η^2 : partial eta squared; * denotes a significant change from baseline to post-treatment in a group, ^a denotes a significant change from baseline to 3-month follow-up in Internet group, # denotes a significant difference between groups at post-treatment.

Table 6.13.

Unadjusted and adjusted descriptive statistics on parent- reported primary and secondary treatment outcomes

	Time	Intervention (n=35)						Control (n=32)					
		Mean	SD	Adjusted Mean	Std Error	95% CI		Mean	SD	Adjusted Mean	Std Error	95% CI	
						Lower	Upper					Lower	Upper
Pediatric Quality of Life Inventory - Physical Health Subscale													
Physical Health	Baseline	32.23	16.07					29.49	18.90				
	Post-treatment	48.93^{*#}	18.96	48.10	2.47	43.16	53.04	24.03	17.42	24.93	2.58	19.77	30.10
	Follow-up	48.57^a	18.34					n/a	n/a				
Wong-Baker FACES Pain Rating Scale													
Pain Intensity	Baseline	5.66	1.83					5.95	2.12				
	Post-treatment	4.69[#]	1.37	4.73	0.25	4.24	5.22	6.91	1.73	6.86	0.26	6.35	7.38
	Follow-up	4.29	1.47					n/a	n/a				
Pediatric Quality of Life Inventory - Psychosocial Subscale													
Psychosocial Health	Baseline	41.43	18.75					36.61	15.15				
	Post-treatment	59.81^{*#}	14.56	58.79	2.08	54.63	62.95	32.14	14.13	33.26	2.18	28.90	37.61
	Follow-up	59.05^a	13.84					n/a	n/a				

SD: standard deviation; η^2 : partial eta squared; * denotes a significant change from T1 to T2 in a group, ^a denotes a significant change from T1 to T3 in Internet group, # denotes a significant difference between groups at T2.

Table 6.13.

Unadjusted and adjusted descriptive statistics on child- reported primary and secondary treatment outcomes (continued).

	Time	Intervention (n=35)					Control (n=32)						
		Mean	SD	Adjusted Mean	Std Error	95% CI		Mean	SD	Adjusted Mean	Std Error	95% CI	
						Lower	Upper					Lower	Upper
Pediatric Quality of Life Inventory - Overall Quality of Life													
Quality of Life Total Score	Baseline	38.23	16.23					34.13	14.04				
	Post-treatment	56.02^{*#}	14.81	54.97	1.99	51.00	58.94	29.31	13.58	30.47	2.08	26.31	34.62
	Follow-up	55.40^a	14.07					n/a	n/a				
Pediatric Pain Coping Inventory													
Pain Coping	Baseline	1.08	0.33					1.08	0.36				
	Post-treatment	1.25^{*#}	0.30	1.26	0.04	1.18	1.33	1.11	0.27	1.11	0.04	1.03	1.18
	Follow-up	1.22^a	0.31					n/a	n/a				
Adults' Response to Children's Symptoms (ARCS) – Protective Behaviour Subscale													
Parental Protectiveness	Baseline	2.09	0.88					2.16	0.84				
	Post-treatment	1.72^{*#}	0.59	1.73	0.09	1.56	1.91	2.08	0.61	2.06	0.09	1.88	2.25
	Follow-up	1.62^a	0.64					n/a	n/a				

SD: standard deviation; η^2 : partial eta squared; * denotes a significant change from T1 to T2 in a group, ^a denotes a significant change from T1 to T3 in Internet group, # denotes a significant difference between groups at T2.

Table 6.13.

Unadjusted and adjusted descriptive statistics on child- reported primary and secondary treatment outcomes (continued).

	Time	Intervention (n=35)					Control (n=32)						
		Mean	SD	Adjusted Mean	Std Error	95% CI	Mean	SD	Adjusted Mean	Std Error	95% CI		
												Lower	Upper
Pain Catastrophizing Scale													
Pain Catastrophizing	Baseline	34.60	12.38					39.09	10.91				
	Post-treatment	24.31^{*#}	9.44	25.22	1.45	22.33	28.11	42.13	10.10	41.13	1.51	38.11	44.16
	Follow-up	23.60^a	9.49					n/a	n/a				
Self-efficacy for Functioning Despite Pain Scale													
Self-efficacy	Baseline	24.49	5.58					26.66	5.27				
	Post-treatment	20.40[#]	4.51	20.69	0.77	19.15	22.24	28.44	4.98	28.12	0.81	26.50	29.73
	Follow-up	20.09	4.82					n/a	n/a				
Numerical Rating Scale													
Sleep	Baseline	6.06	2.54					7.19	2.63				
	Post-treatment	4.34^{*#}	1.64	4.65	0.33	3.99	5.31	7.31	3.03	6.99	0.35	6.29	7.67
	Follow-up	4.46^a	1.62					n/a	n/a				

SD: standard deviation; η^2 : partial eta squared; * denotes a significant change from T1 to T2 in a group, ^adenotes a significant change from T1 to T3 in Internet group, # denotes a significant difference between groups at T2.

6.4.21 Maintenance of Treatment Effects at Three Month Follow-up

One-way ANOVAs with repeated measures were conducted to evaluate the maintenance of treatment effects at 3-month follow-up for the intervention group on retrospective report of clinical outcomes. All analyses were conducted with the intent-to-treat sample using the last observation carried forward approach and Bonferroni contrasts.

6.4.22 Physical health

Child retrospective report of physical health significantly improved in the Internet intervention group, Wilks' Lambda = .82, overall $F(2, 65) = 7.01$, $p = .002$. Pairwise comparisons showed that treatment effects were significantly different at T1–T2 and T1–T3 ($p = .001$). Comparisons at T2–T3 did not differ significantly ($p = .70$) suggesting these effects were maintained at follow-up. This effect was large, partial $\eta^2 = .18$.

Similarly, parent retrospective report of child physical health significantly improved in the Internet intervention group, Wilks' Lambda = .91, overall $F(2, 65) = 3.33$, $p = .04$. Pairwise comparisons showed that treatment effects were significantly different at T1–T2 and T1–T3 ($p = .04$). Comparisons at T2–T3 did not differ significantly ($p = .99$) suggesting these effects were maintained at follow-up. This effect was medium, partial $\eta^2 = .09$.

6.4.23 Pain intensity

Child retrospective report of mean pain intensity improved in the Internet intervention group, however these effects were not significant, Wilks' Lambda = .94, overall $F(2, 65) = 2.21$, $p = .12$. Pairwise comparisons showed that treatment effects did not differ significantly at T1–T2 ($p = .12$), T1–T3 ($p = .14$) or T2–T3 ($p = .99$).

Similarly, parent retrospective report of child pain intensity improved in the Internet intervention group, however these effects were not

significant, Wilks' Lambda = .92, overall $F(2, 65) = 2.81, p = .07$. Pairwise comparisons showed that treatment effects did not differ significantly at T1–T2 and T1–T3 ($p = .99$) or T2–T3 ($p = .06$).

6.4.24 Psychosocial health

Analysis of child retrospective report of psychosocial health showed a significant main effect for time in the Internet intervention group, Wilks' Lambda = .91, overall $F(2, 65) = 3.29, p = .04$. However, pairwise comparisons showed that treatment effects did not differ significantly at T1–T2 ($p = .07$), T1–T3 ($p = .18$) or T2–T3 ($p = .78$).

Parent retrospective report of child psychosocial health significantly improved in the Internet intervention group, Wilks' Lambda = .87, overall $F(2, 65) = 5.03, p = .009$. In contrast with child report, pairwise comparisons showed that treatment effects were significantly different at T1–T2 ($p = .007$) and T1–T3 ($p = .009$). Comparisons at T2–T3 did not differ significantly ($p = .99$) suggesting these effects were maintained at follow-up. This effect was medium, partial $\eta^2 = .13$.

6.4.25 Total quality of life

Child retrospective report of total quality of life significantly improved in the Internet intervention group, Wilks' Lambda = .88, overall $F(2, 65) = 4.44, p = .02$. Pairwise comparisons showed that treatment effects were significantly different at T1–T2 ($p = .013$) and T1–T3 significant ($p = .024$). Comparisons at T2–T3 did not differ significantly ($p = .99$) suggesting these effects were maintained at follow-up. This effect was medium, partial $\eta^2 = .12$.

Similarly, parent retrospective report of child total quality of life significantly improved in the treatment group, Wilks' Lambda = .86, overall $F(2, 65) = 5.18, p = .008$. Pairwise comparisons showed that treatment effects were significantly different at T1–T2 ($p = .006$) and T1–T3 ($p = .007$).

Comparisons at T2-T3 did not differ significantly ($p = .99$) suggesting these effects were maintained at follow-up. This effect was large, partial $\eta^2 = .14$.

6.4.26 Pain coping

Child retrospective report of pain coping significantly improved in the Internet intervention group, Wilks' Lambda = .89, overall $F(2, 65) = 3.90$, $p = .025$. Pairwise comparisons showed that treatment effects were significantly different at T1-T2 ($p = .02$) and T1-T3 ($p = .04$). Comparisons at T2-T3 did not differ significantly ($p = .99$) suggesting these effects were maintained at follow-up. This effect was medium, partial $\eta^2 = .11$.

Parent retrospective report of child pain coping significantly improved in the Internet intervention group Wilks' Lambda = .87, overall $F(2, 65) = 4.94$, $p = .01$. Pairwise comparisons showed that treatment effects were significantly different at T1-T2 ($p = .007$) and T1-T3 ($p = 0.013$). Comparisons at T2-T3 did not differ significantly ($p = .34$) suggesting these effects were maintained at follow-up. This effect was large, with partial $\eta^2 = .13$.

6.4.27 Sleep

Child retrospective report of sleep quality significantly improved in the Internet intervention group, Wilks' Lambda = .91, overall $F(2, 65) = 3.27$, $p = .044$). Pairwise comparisons showed that treatment effects were significantly different at T1-T2 ($p = .044$) and T1-T3 ($p = .038$). Comparisons at T2-T3 did not differ significantly ($p = .99$) suggesting these effects were maintained at follow-up. This effect was medium, partial $\eta^2 = .09$.

Parent retrospective report of child sleep quality significantly improved in the Internet intervention group, Wilks' Lambda = .88, overall $F(2, 65) = 4.26$, $p = .018$. Pairwise comparisons showed that treatment effects were significantly different at T1-T2 ($p = .017$) and T1-T3 ($p =$

.034). Comparisons at T2-T3 did not differ significantly ($p = .99$) suggesting these effects were maintained at follow-up. This effect was medium, partial $\eta^2 = .12$.

Self-efficacy

Child retrospective report of self-efficacy significantly improved in the Internet intervention group, Wilks' Lambda = .88, overall $F(2, 65) = 4.52, p = .014$. Pairwise comparisons showed that treatment effects were significantly different at T1-T2 ($p = .011$) and T1-T3 ($p = .021$). Comparisons at T2-T3 did not differ significantly ($p = .92$) suggesting these effects were maintained at follow-up. This effect was medium, partial $\eta^2 = .12$.

In contrast, parent retrospective report of child self-efficacy improved in the Internet intervention group, however these effects were not significant, Wilks' Lambda = .95, overall $F(2, 65) = 1.79, p = .18$. Pairwise comparisons showed that treatment effects did not differ significantly at T1-T2 ($p = .35$), T1-T3 ($p = .21$) or T2-T3 ($p = .99$).

6.4.28 Pain catastrophising

Child retrospective report of pain catastrophising significantly improved in the Internet intervention group, Wilks' Lambda = .79, overall $F(2, 65) = 8.83, p < .001$. Pairwise comparisons showed that treatment effects were significantly different at T1-T2 and T1-T3 ($< .001$). Comparisons at T2-T3 did not differ significantly ($p = .99$) suggesting these effects were maintained at follow-up. This effect was large, partial $\eta^2 = .21$.

Parent retrospective report of child pain catastrophising significantly improved in the Internet intervention group, Wilks' Lambda = .91, overall $F(2, 65) = 3.44, p = .04$. Pairwise comparisons showed that treatment effects were significantly different at T1-T2 ($p = .04$) and T1-T3 ($p = .03$). Comparisons at T2-T3 did not differ significantly ($p = .99$) suggesting these effects were maintained at follow-up. This effect was medium, partial $\eta^2 =$

.10.

6.4.29 Parent protectiveness

Child retrospective report of parental protective behaviour improved in the treatment group, however these effects were not significant, Wilks' Lambda = .93, overall $F(2, 65) = 2.28$, $p = 0.11$. Pairwise comparisons showed that treatment effects did not differ significantly at T1–T2 ($p = 0.31$), T1–T3 ($p = 0.26$) or T2–T3 ($p = .36$).

In contrast, parent retrospective report of parental protective behaviour significantly improved in the Internet intervention group, Wilks' Lambda = .86, overall $F(2, 65) = 5.46$, $p = .006$. Pairwise comparisons showed that treatment effects were significantly different at T1–T2 ($p = .045$) and T1–T3 ($p = .01$). Comparisons at T2–T3 did not differ significantly ($p = .21$) suggesting these effects were maintained at follow-up. This effect was large, partial $\eta^2 = .14$.

6.4.30 Adverse events

No adverse events were reported by any of the participants who took part in this study.

6.4.31 Intervention Mapping

Usage data and qualitative feedback suggest that the child participants who took part in the Internet intervention, seemed to accept the value of adaptive coping behaviour (PO.1, PO.9) and used weekly goal setting (PO.7, PO.12). Child participants also used action planning strategies (PO.5, PO.6, PO.8, PO.11) to identify how coping sub-behaviours could be improved (see Table 6.14). The weekly check-in pages encouraged these motivated participants to monitor their coping behaviour and set rewards (PO.6, PO.7). Objective usage data in addition to self-report data, show children set graded goals for themselves and accessed video and

audio materials teaching adaptive coping strategies e.g. guided visualisation (PO.9, PO.12, PO.13). Input on the interactive, text-based strategies e.g. problem solving, BEST communication techniques suggest some children tried to apply CBT strategies for better functioning in school and social situations (PO.10, PO.11, PO.13). Finally, child participants were proactive in selecting their preferred CBT strategies (PO.8, PO.13). It should be noted that usage data and qualitative feedback suggest the completion of CBT strategies was largely based on personal preference and not necessarily support needs. Less popular strategies were either incomplete or skipped over, suggesting appropriate CBT strategies may not necessarily have been selected.

Table 6.14

Desired change and performance objectives of the Feeling Better intervention

Change objective 1:	Enhance participants self-efficacy by end of programme and 3-month follow-up
Performance objective 1	Children accept the importance of pain SM
Performance objective 2	Children become aware of current pain SM behaviour
Performance objective 3	Children identify barriers and facilitators to be addressed
Performance objective 4	Children identify ACBs (sub-behaviours) to be improved
Performance objective 5	Children understand how and when to use CBT strategies to cope with pain
Performance objective 6	Children understand how to monitor progress in ACBs (sub-behaviours)
Performance objective 7	Children identify realistic goals: SMART goals
Performance objective 8	Children decide to use relevant CBT strategies to manage pain
Change objective 2:	Increase participants' use of CBT strategies by end of programme and 3-month follow-up
Performance objective 9	Children cope with barriers and use resources
Performance objective 10	Children select appropriate CBT strategies to support functioning despite pain
Performance objective 11	Children seek social support
Performance objective 12	Children work toward graded, realistic goals
Performance objective 13	Children use self-regulation strategies to support practice and recall skills training

ACB: adaptive coping behaviour(s); CBT: cognitive behavioural therapy; SM: self-management; SMART: Specific, Measurable, Attainable, Realistic and Timely;

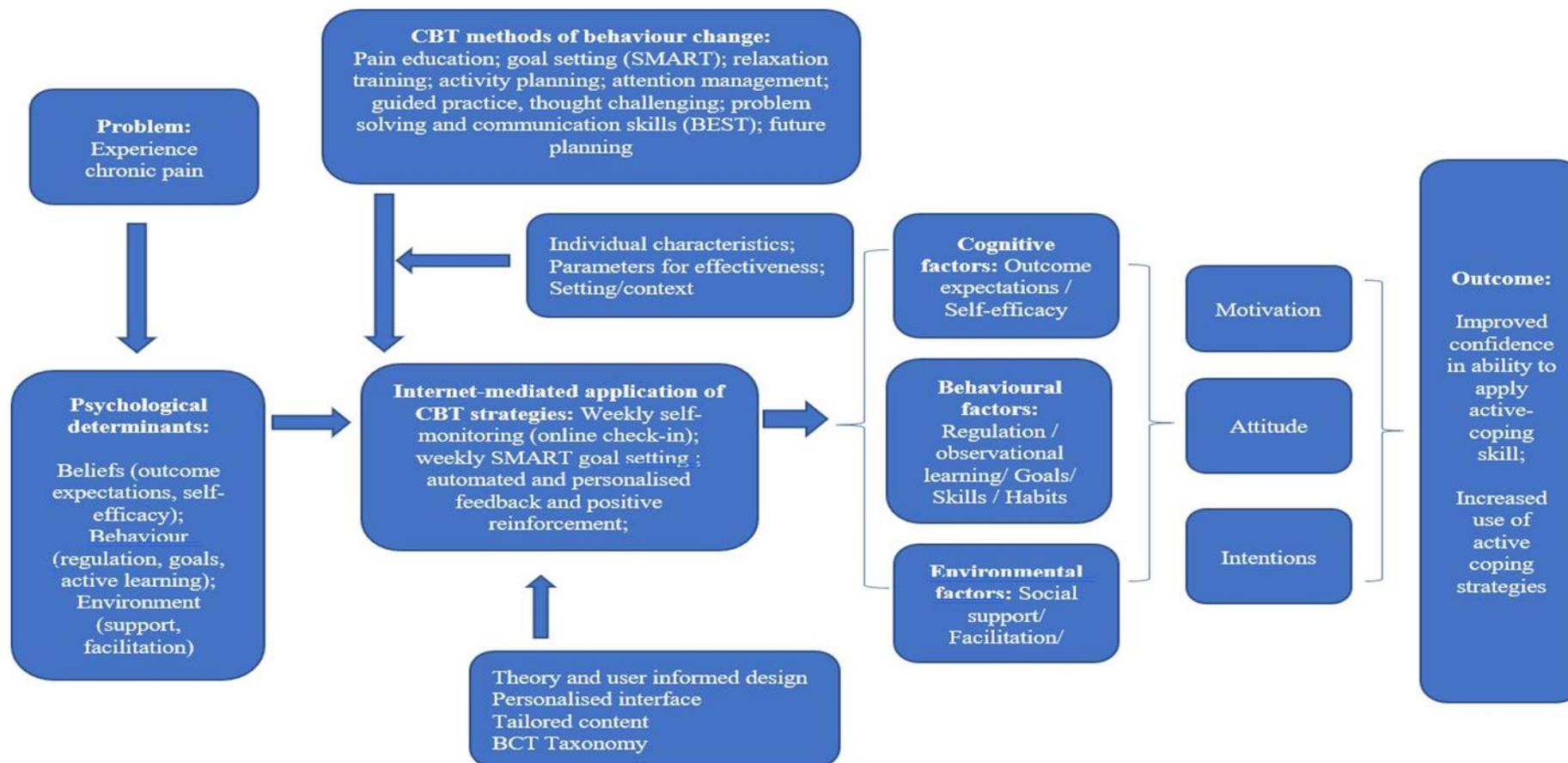


Figure 6.4 Logic Model developed for the *Feeling Better* intervention

6.5 Discussion

The primary aim of this research was to establish the feasibility and acceptability of the *Feeling Better* pain management programme, delivered remotely and adapted for pre- adolescent children with chronic pain and their care-givers. Feasibility evaluations were based recruitment, retention, adherence, satisfaction and usability. A secondary aim of this research was to evaluate the potential effectiveness of the *Feeling Better* programme. Efficacy evaluations focused on change from baseline in several core outcomes including physical health (functional limitations), pain intensity and psychosocial health (mood, social and role functioning), pain catastrophising, coping strategies and pain- related self-efficacy. It was hypothesised that participants pain intensity and interference, pain self-efficacy, pain catastrophising and use of coping strategies would be improved because of the *Feeling Better* intervention.

6.5.1 Overall findings

With respect to feasibility, the adapted pain management programme was found to be feasible and acceptable to child and parent participants. This evaluation of the *Feeling Better* pain management programme suggests that a follow-up randomised controlled trial with a larger sample size may be feasible and warranted. Recruitment targets were achieved, and attrition rates were comparable to other published CBT-based, online interventions (Hicks et al., 2006; Palermo et al., 2009; Stinson et al., 2010; Fisher et al., 2015). The *Feeling Better* programme was associated with greater improvement in self-reported clinical outcomes compared to the waitlist control group.

As per child report, the beneficial effects were maintained for several outcomes at three- month follow-up (physical health, total quality of life, pain coping, self-efficacy, pain catastrophising, sleep). Several other key outcomes (pain intensity, pain catastrophising, parental protectiveness and psychosocial health) favoured the Internet intervention but were not statistically significant at three-month follow-up. Follow-up analyses

included only 10 participants and could not be compared across trial arms. This study was not powered to detect clinically meaningful changes and a larger trial and longer follow-up period is required to establish if the online *Feeling Better* programme may be an effective means of enhancing active pain self-management in this group. Overall, the findings from the present study are consistent with other Internet-based pain management interventions and are discussed in further detail below.

6.5.2 Recruitment

It took 12 months to recruit participants to the feasibility trial. Initial response to recruitment was poorer than expected, therefore, early in the recruitment process, it was decided that inclusion criteria should be expanded to include participants outside the Republic of Ireland. Following this decision, the minimum target for randomisation of approximately 59 participants was exceeded. The recruitment process was streamlined and participants who expressed an online interest in the study were taken directly to the intervention website where they had access to the participant information forms and initial screening questions. There was no reason to leave the *Feeling Better* website as the randomisation was automated and a password for the *Feeling Better* programme issued upon completion of the baseline survey and randomisation process. The lack of delay in carrying out these procedures directly contributed to the high conversion from initial enquiry to registration.

Despite this success, recruitment from the community was challenging because of the nature of the study and the target sample. Children with chronic pain are a vulnerable population and therefore naturally difficult to access. At the time of recruitment, there were also several other research teams attempting to access the same age-specific chronic pain population. In addition, internet and technology mediated interventions for this group have not previously been carried out in the Republic of Ireland to the author's knowledge and the novelty of the approach may have been a deterrent for some. The recruitment process itself

was challenging for two reasons, the first being the workload and time commitment required to maintain a presence on condition-specific social media pages and platforms (e.g. the Irish Children's Arthritis Network and Facebook). Lack of social media training and budget constraints undoubtedly contributed to inefficiency of labour. Someone with expertise in social media marketing might have promoted the study more effectively.

6.5.3 Retention

Although comparable with similar interventions, the attrition rate for the intervention phase in the current study was relatively high (28%) (Hicks et al., 2006; Rapoff et al., 2014; Voerman et al., 2015). The difference between this study and similar interventions is that the current study recruited entirely from the community and via social media. As observed in our systematic review, almost all similar interventions have tended to recruit from within a hospital or clinic setting. For example, a similarly designed trial targeting adolescents (Palermo et al., 2009) used face to face recruitment from a clinic setting over a two-year period and experienced low attrition rates (8%) compared to the 28% reported in the *Feeling Better* trial that relied on social media as the primary source of recruitment. In this field of research, only one other trial has recruited solely from the community (Trautmann & Kröner-Herwig, 2010) and a second trial has recruited from both the community and through advertising in outpatient clinics (Hicks 2006). Trautmann & Kröner-Herwig (2010) also report relatively high withdrawal (25/45 participants, CBT group: 13/24) whereas Hicks et al., (2006) report a lower rate (15/47, CBT group 7/25 participants) using a combined approach. It is possible face to face recruitment from a clinic setting may lend a certain credibility and accountability to the recruitment process that promotes adherence, and which is not observed when reliant on social media.

It is possible that assessment fatigue contributed to the loss to follow-up. In response to reminders to complete post-treatment and follow-up assessment, some participants offered feedback suggesting the

assessment measures were extensive. Further piloting of assessment procedures may be necessary to accurately inform a larger trial. Dropout was higher in the waitlist control arm of the study. This may be due to the lack of contact throughout the active phase of the intervention (i.e. 9-week intervention period). Parent participants in the WLC expressed an understanding but also disappointment at group allocation particularly when explaining this to their child. This suggests further counselling at baseline or additional methods i.e. visual illustrations of the randomisation process might to be usefully applied when explaining the randomisation process to young people and thereby reduce disappointment or misunderstanding. Participants who engaged with the intervention cited tailored and personalized feedback as the main appeal of the programme. The degree of personalized, automated content i.e. messages of support and encouragement and tailored weekly progress reports were viewed as a significant advantage. This is consistent with research that suggests personalised feedback is associated with increased intervention use and reduced attrition (Morrison et al., 2012).

6.5.4 Adherence

All 35 dyads allocated to the Internet intervention logged onto the website at least once. This figure supports the early decision to embed the assessment methods in the host website which delivered the psychological intervention and thereby reduce pre-enrolment attrition. A total of 26 (74%) children completed the programme, these figures are comparable with similar pain self-management interventions (Palermo et al., 2009) and meet pre-determined feasibility criteria. There is no established, optimal dose-response relationship for online psychological interventions to provide guidance about the amount of intervention exposure most likely to be efficacious. A systematic review by Carnes et al (2011) recommend self-management programmes of approximately 8 weeks duration for optimal benefit (Carnes et al., 2012). The criteria for programme completion was 5 or more modules which take approximately 20 minutes to complete. The

average number of unique logins per child participant allocated to the Internet intervention was 14 and the average number of unique logins among ‘completers’ was 18. Also, that average time taken to complete a module was 24 minutes. It may be that the intensity of the programme was too much for this age group.

The nine-week *Feeling Better* programme was completed in approximately 11 weeks on average by most child participants. Progress was slow but continuous. Reasons for delay included pain flare ups and family commitments. This is consistent with research that suggests children who participate in Internet-mediated treatment, progress more slowly through treatment sessions (March et al., 2009). This is also consistent with existing 8-week Internet-based interventions for adolescent chronic pain management that report the treatment completion takes approximately 8-10 weeks (Palermo et al., 2009; Law, Beals-Erickson, Noel, Claar, & Palermo, 2015; Palermo et al., 2016).

Treatment compliance data should be interpreted with caution due to the measures of treatment and homework compliance used. Like existing Internet interventions, in this field, the current trial evaluated treatment compliance in terms of the number of modules (i.e. treatment sessions) completed and homework tasks submitted (i.e. homework compliance). This method alone does not account for the accuracy with which tasks were completed or the increasing complexity of CBT strategies as treatment progresses. A measure of treatment compliance based on the weight of a strategy i.e. complexity of the task or on the accuracy with which the strategy was completed may produce a more conservative estimate of treatment and homework compliance. In the current trial, participants were considered ‘completers’ if they completed 5 or more modules. However, in line with CBT practice, the complexity of treatment increased sequentially with more sophisticated CBT strategies introduced in the second half of the programme e.g. Sessions 6, 7 and 8 featured more sophisticated cognitive re-structuring, problem solving and communication skills training. Thus, using only quantitative measures of compliance, i.e. session compliance and homework compliance, some participants may be considered ‘completers’

without having attempted the more complex CBT strategies.

Also, it may be important to incorporate qualitative assessment of compliance in trials such as this where the clinical content was adapted from material designed for older individuals. Accuracy of task completion indicates that the instructions for task completion and the function of the task were understood and potentially internalised by participants. This could be a better measure of the success of the adaptation process and specifically the ease of understanding. Future iterations of this trial might consider a comparison of quantitative and qualitative compliance measures to more accurately capture compliance.

6.5.5 Qualitative feedback

6.5.6 Facilitators and barriers to engagement

Barriers to engagement for parent participants were competing life demands and among those parents with older children (ages 8-12) a lack of need of parent support to complete the programme. Barriers to engagement cited by children were lack of access to peers, time commitment and difficulty understanding content. Barriers to engagement cited by parents of younger children (5-7 years) include programme intensity and lack of access to peers.

Suggestions for improvement related to peer interaction and access to health professionals, and integration with usual medical care. Given the high degree of current interest in the development of digital health interventions, it is important to explore how to better engage young children with chronic conditions. Future interventions should continue to focus on the facilitators which promote adherence among participants with chronic conditions to support active self-management. The present study found that tutor support, the focus on personal goals and monitored progress toward goals were the most commonly cited facilitators of engagement, followed by automated positive feedback (positive reinforcement messages). This is consistent with research that shows that regardless of treatment condition the presence of therapist support may be associated with positive, non-specific

effects including significant increases in the use of adaptive pain coping strategies, pain coping efficacy and lower catastrophizing (Kashikar- Zuck et al., 2012a). The question of what type of support and how much is optimal in e-Health is an important and unanswered question (McGuire, Henderson & McGrath, 2017). Participant preferences for CBT-strategies and design features may be based on the age-appropriateness of the strategy i.e. the cognitive and emotional skill level of a child at 5 years of age is very different to that of a 12-year-old. Alternatively, these preferences may be based on the child's comfort level addressing emotional issues or making cognitive-emotional connections i.e. linking thoughts to emotions and behaviour. Comparisons across sub-groups of 'completers' in terms of usage was not possible due to small sample size and lack of diversity in terms of participant age, gender or pain condition.

6.5.7 Intervention mapping

As stated in Chapter 2, an intervention mapping approach was used to guide the design and development of this research. According to usage data and qualitative input in the *Feeling Better* intervention, the behavioural outcomes and performance objectives identified at the outset were largely achieved. This supports the utility of an intervention mapping protocol in each phase of the study.

6.5.8 Involving parents

Usage data indicated that parents made little use of their specifically designed pathway through the programme. This may mean that they found their parent pathway and/or the treatment approach to be less acceptable than the usability research had previously indicated. While usage data suggests parents were not involved in the treatment approach, qualitative feedback from children and parents suggests they were more involved. Response to open-ended treatment evaluation questions suggested parents preferred to look over their child's shoulder during sessions, opting to

complete the programme as a team, using only the child pathway through the programme. In other words, parents preferred to act as “Coach” based on the information gathered in the child section rather than the more detailed and parallel, information-only sessions set out for their use. Overall, qualitative feedback from children and parents was positive regarding the involvement of parents. These findings are in line with Palermo et al., (2009) who conducted a similar study in terms of trial design (i.e. two-armed, waitlist controlled) and programme intensity (i.e. 8 modules, multiple pain conditions and behaviours targeted). The research team evaluated the efficacy of Internet-based CBT for adolescents with chronic pain and their parents. The authors observed a lower completion rate by parents compared to their adolescent children (Palermo et al., 2009). However, the completion rate in the *Feeling Better* trial was still lower, suggesting parents were not effectively involved in the treatment programme. For this age group, specifically, it would be helpful to determine the optimal extent of parental involvement. As might be expected, younger children are less-independent users compared to adolescents. Further research is needed to determine an acceptable level of parental involvement for parents of pre-adolescent children.

6.5.9 Consideration of study design and clinical outcomes

Comparison of intervention design and clinical outcomes across Internet-based interventions for chronic pain highlight several methodological and operational differences. Internet-based interventions for paediatric pain typically implement a randomised controlled trial design, most are pilot trials with one, high quality exception (Palermo et al., 2016). In line with the present study, all interventions provided positive feedback during the active phase of the intervention. Minimal tutor (therapist) support was delivered using additional telephone and email correspondence (Hicks et al., 2006; Palermo et al., 2009; Palermo et al., 2015; Palermo et al., 2016). All interventions were based on CBT principles. Some of the interventions did not explicitly report the application of CBT but described the clinical

content of the intervention as consistent with this approach. In contrast with the current study, most interventions consisted of clinical populations recruited from secondary care clinics or research centres (Cottrell et al., 2007; Palermo et al., 2009; Stinson et. al., 2010; Rapoff et al., 2014; Law et al., 2015; Palermo et al., 2016), only one trial recruited from the community (Trautmann & Kröner-Herwig, 2010). Data collection also differed between interventions, some interventions used online questionnaires (Law et al., 2015; Palermo et al., 2016) others used printed materials (Connelly et al., 2006; Palermo et al., 2009). Finally, some interventions focused on only one chronic pain condition such as arthritis (Stinson et. al., 2010a), chronic headache (Law et al., 2015) or mixed pain conditions (Palermo et al., 2009; Palermo et al., 2016). These differences between interventions are important as they may account for contrasting findings between treatment studies for paediatric chronic pain.

Trautmann and colleagues (2010) conducted an Internet-based RCT of CBT in comparison with applied relaxation and education-based control conditions for children and adolescents (ages 10-18 years) with recurrent headache (Trautmann & Kröner-Herwig, 2010). Also using the PC-S, the authors found improvement in pain catastrophising across all groups but no significant between-group differences in pain catastrophising at post-treatment. However important improvements were noted in the CBT group over time (6 months post- treatment). This contrasts with the current study which found between group differences in catastrophising but no maintenance of beneficial effect at follow-up as per child report. It may be the observed decrease in pain catastrophising was a non-specific effect of treatment (i.e. placebo) rather than specific response to the online *Feeling Better* programme. Alternatively, this contrast in findings may be explained by differing samples. The current systematic review (Chapter 3) and comparative evaluations of efficacy suggest greater beneficial effects in response to Internet-based CBT among children with chronic headache in comparison with mixed pain conditions.

The current study observed a significant pre-post reduction in pain intensity, however this was not maintained at three-month follow-up. This

finding is in contrast with a similarly designed RCT of an Internet intervention which examines the efficacy of CBT compared to a waitlist control condition for children (9-16 years) with recurrent abdominal pain or headache. Hicks and colleagues (2006) found a significant improvement in pain intensity at one month and three - month follow-up. The authors reported mild to moderate pain severity at baseline (4.6/10) in comparison with the moderate pain severity (6.3/10) reported in the current study. It may be that these treatment effects do not generalise to more severe pain populations and specifically the maintenance of beneficial effects may not be observed after a brief intervention for those with more severe pain at baseline. It may be that the time-frame was too short to observe clinically meaningful effects given the profile of participants who completed the programme. The current sample was comprised largely of children with arthritic pain. Trudeau et al., (2015) suggested that patients with arthritic pain may need longer than six months before change in self-efficacy and pain catastrophising influence self-reported pain intensity or severity in a clinically meaningful way.

As hypothesised, there was a significant reduction in child and parent reported parental protectiveness at post-treatment in the intervention group compared to the waitlist control condition. However, unlike similar interventions (Palermo et al., 2009; Palermo et al., 2016), these effects were not maintained at follow-up. Contrasting findings may be explained by differences in key study parameters. It may also be that the negative impact of over-protective behaviour is more pronounced in adolescents compared to pre-adolescent children. Further research is required to determine if the degree of protective behaviour is a mediator of treatment specific effects. In summary the findings from the current study have demonstrated an online version of the *Feeling Better* programme is both feasible and may facilitate significant reduction in pain-related clinical outcomes.

6.5.10 Strengths

This study had several strengths. First, the *Feeling Better*

programme focused on pre- adolescent children with chronic pain. This is an under-researched, poorly understood population particularly in the context of Internet intervention development. Unlike similar Internet interventions the age limit was focused on children between 5-12 years of age. This is important as our systematic review found only two studies (Connelly et al., 2006; Hicks et al., 2006) that included children in this age range. These analyses were not stratified according to age group therefore we cannot draw comparisons.

This information contributes to current understanding of the acceptability, wider applicability and relevance of Internet-delivered interventions for pre-adolescent paediatric populations. The *Feeling Better* programme involved parents in their children's treatment. Previous research has shown the importance of family-focused cognitive behavioural therapy in the successful treatment of child and adolescent chronic conditions (Goubert, Eccleston, Vervoort, Jordan, & Crombez, 2006; Palermo et al., 2009; Fales, Essner, Harris, & Palermo, 2014; Palermo et al., 2016).

This trial evaluated intervention effects on a range of PedIMPACT recommended core outcome domains including physical functioning, pain, emotional functioning, treatment satisfaction and parental protective behaviour (McGrath et al., 2008).

A high participation rate was observed among those who contacted the research team to enquire about the programme (95% of eligible children who initially expressed an interest subsequently accessed the study) indicating a high acceptance of internet-based psychological treatment delivery among this population. According to qualitative feedback from parents and children, the virtual incentive (i.e. social reward: receive a promotion and move up the ranks from powder monkey to captain with every module completed), the focus on goal setting, tracking and achievement and access to an online tutor were the most engaging features of the programme. Computer game-like incentives and terminology may appeal to this cohort. Future design considerations might explore the effect of other game design features (e.g. power-up when you complete a CBT strategy, lose rank when you fail to complete homework) on participant

engagement. High completion rate may also be explained by the commitment we asked all participants to make as part of the Pain Education module. Asking participants to choose to “join the crew” and make that commitment may have inspired greater adherence than would have been observed otherwise (Pereles, Lockyer, Hogan, Gondocz, & Parboosingh, 1997).

Commitment to the programme may be the result of a successfully personalising the programme and incorporating clinical content that aligns with the needs and preferences of children with chronic pain associated with JIA (Study 2). The topics chosen were based on the literature (Tong et al., 2012; McManus et al., 2014) and feedback from a preceding qualitative study involving children and parents with JIA (see Chapter 3). Many of the children who participated in the study had JIA indicating the *Feeling Better* programme may have been optimised for their preferences. Another reason for the relatively high completion rates may be the number of interactive and personalised mode of delivery features throughout each of the eight modules tested. The *Feeling Better* programme may have engaged participants because it is based on an existing theory with a focus on self-management and social facilitation. Tutor support was highlighted by participants as a significant strength of the study. This is consistent with research which suggests Internet-based psychological therapies with additional tutor or therapist support are more beneficial than those without (SPEK et al., 2007). Finally, randomisation occurred immediately after baseline data collection and was conducted using a third party, automated system. However, due to the nature of the intervention it was not possible to blind the researcher to allocation.

6.5.11 Limitations and future research directions

This study has several limitations. First, this study was limited by a small sample size which affects the confidence with which we can interpret these results. As shown in the systematic review reported in Chapter 3 and recent topical reviews, this is a typical problem in the

psychological, paediatric intervention studies (Kashikar-Zuck, 2010; Morley & Williams, 2015). Reasons for low sample size might include the necessary challenges associated with access to eligible participants and the anonymity and lack of accountability associated with digital health interventions.

Second, the current design did not allow the separation of treatment from placebo effects or inferences regarding possible long-term effects of treatment. The absence of a waitlist control condition at three-month follow-up makes it difficult to determine the extent to which positive findings for the *Feeling Better* Internet intervention from post-treatment to three-month follow-up represent the effects of treatment. Thus, this data should be interpreted with caution. Future studies should use a comparative attention control condition which would address these issues. This study would have been strengthened by the inclusion of a clinic-based treatment comparison condition to determine the efficacy of Internet-based treatment compared to clinic-based interventions for pre-adolescent children within this age group. This was beyond the scope and resources of the current study. The results of this study demonstrate medium to large effect sizes for Internet-based CBT for paediatric chronic pain management. This is like existing clinic-based studies suggesting that the online *Feeling Better* programme warrants further investigation as a potential adjunct to clinic-based treatment or source of support for those awaiting access to psychological services.

Third, despite the exploratory analyses conducted, this trial was not powered to detect the influence of mediating and moderating factors on the included outcomes. This is an area warranting further investigation as little is known about individual differences in treatment response in paediatric chronic pain trials (Eccleston et al., 2014a).

Fourth, despite recruiting from the community and opening the study to participants outside of Ireland, the current sample comprised of an extremely homogenous group (Caucasian and well educated). This diminishes confidence in the representativeness of the sample and generalisability of results to a wider population. Moreover, this study was

not conducted in a ‘real-world’ clinical context. It may be that participants recruited to the present study suffer with milder pain conditions than those observed in clinical populations.

Fifth, feasibility was established but the recruitment challenges experienced suggest the approach to recruitment could be greatly improved. Recruitment via social media was very time consuming and labour intensive as many of the parent-led networks on Facebook are closed groups and permission was required to promote this research. Also, strict rules had to be followed in terms of how many times and how often this research could be promoted on given Facebook pages. To keep the study information in the current to recent news feed that it might be viewed by the maximum number of people the process of releasing information feeds, summaries and briefs explaining this project were issued repeatedly. In addition, there was a lack of available information from social media platforms about the level of interest in the study. For example, information about the *Feeling Better* trial was posted to many pain-related charity and support group pages on Facebook. Without access to the usage data associated with these pages it is impossible to ascertain how many individuals read this summary information and decided not to proceed further.

Challenges also included lack of incentives for patients to participate, onerous informed consent and assent procedures, and other end-user issues such as lack of skills or confidence using a computer. The recruitment process was streamlined to facilitate online recruitment and baseline assessment with or without researcher assistance. However, the parent-focused recruitment approach employed resulted in several incomplete online surveys where the parent section was submitted but the child section remained blank. This suggests the current recruitment methods and information materials successfully targeted parents but did not appeal to young people. Fourth, to avoid over-assessing participants, the influence of treatment expectancy was explored using an anchored, visual analogue scale rather than an additional and validated treatment expectancy questionnaire. Future studies might revise this decision.

Sixth, external validity may have been compromised by the

inclusion criteria based on contextual circumstances for example, that all participants must have regular access to a computer and the internet. Individuals who were otherwise eligible to take part may have been excluded based on these criteria.

Finally, there are several issues that warrant further investigation in relation to the *Feeling Better* programme. First it is important to examine whether the email communications are necessary component of the *Feeling Better* intervention and whether equivalent efficacy would be demonstrated in an unsupported intervention. Second, it is important to investigate the economic viability of Internet-based intervention in terms of how they compare to the overall costs associated with traditional therapy.

6.5.12 Conclusion

Feeling Better is the first randomised controlled trial to evaluate the feasibility and potential efficacy of cognitive behaviour therapy designed specifically for pre-adolescent chronic pain in an online setting. This feasibility study shows that the online *Feeling Better* intervention warrants further evaluation in a large scale, pilot randomised controlled trial, and that it is feasible to do so. These findings support the cognitive behavioural model of chronic pain. The impact of this brief CBT intervention on patient outcomes beyond those associated with the waitlist control group indicates the promise of the *Feeling Better* intervention for children with chronic pain. The next logical step would be a more rigorous research design, involving a comparative attention control in terms of participant commitment and treatment delivery. Future research should evaluate the true impact of the *Feeling Better* programme on clinical psychological and behavioural outcomes. Also, more advanced statistical techniques might be employed in future trials to explore the reciprocal effect of one variable on another (i.e. Mediation analyses), specifically with respect to age and compliance (session and homework) in relation to treatment outcomes. How the data fits with the intervention matrix linking

hypothesised SCT constructs to chronic pain outcomes might also be explored using advanced techniques (i.e. Structural Equation Modelling). Finally, it should be noted that the current study was developed with feedback from children with various forms of JIA and EDS and the sample of participants who took part in the feasibility trial were largely children with various subtypes of JIA and musculoskeletal conditions. It may be that the *Feeling Better* programme was particularly tailored to the needs of this group. Future research might consider tailoring the Feeling Better intervention to patient subgroups categorised by level of pain, function or distress. A definitive evaluation of the effectiveness of *Feeling Better* programme in a large scale RCT should involve a multi-centre recruitment approach and longer follow-up periods.

Chapter 7 Concluding comments

7.1 Chapter overview

This chapter will present a summary of the overall findings of this research and contribution made by each study to the adaptation, development and evaluation of an online version of the *Feeling Better* pain management programme for pre-adolescent children with chronic pain and their care-givers. Research aims, and objectives are reviewed alongside the main findings from each study. This is followed by a summary of the key contributions and implications of the findings for future research and practice. The limitations of each study will be described and directions for future research will be suggested. Finally, this chapter will close with a summary of overall conclusions.

7.2 Summary of overall research findings

In Study 1 we evaluated the effectiveness of psychological interventions for paediatric chronic pain delivered using information and communication technology (RQ1). The systematic review did not fully answer the research question due to a lack of data and poor standards of trial report. However, the potential of ICT-based therapies was established. The results from Study 1 provide tentative evidence of effectiveness of existing, ICT-based psychological therapies for paediatric chronic headache pain post-treatment. Also, treatment satisfaction was significantly higher in the treatment group compared to the control group suggesting this mode of delivery may be an acceptable solution to current resource and access issues. These results also supported findings from the wider cognitive-clinical literature suggesting CBT is the most researched and robust psychological treatment for chronic pain to date (Morley, 2011; Morley & Williams, 2015). Despite the body of research supporting the use of CBT, the mechanisms by which it leads to improved pain self-management and outcomes remain unclear. Explicit account of the hypothesised mechanism of behaviour change was lacking in each of the included interventions.

Retrospective, theoretical content analysis identified common and

potentially important theoretical constructs targeted by the included interventions, namely: knowledge, skills, behaviour regulation, memory, attention and decision processes, social influences and beliefs about consequences and capabilities. Less frequently reported, but potentially important constructs were also identified: goals, reinforcement and emotion. According to content analyses, social identity, intentions and optimism were not targeted by any of the included interventions. Interventions that excluded the BCT 6.1 ‘demonstration of the behaviour’ was associated with greater estimates of effect in response to treatment compared to interventions that were perceived to have incorporated this technique. Subgroup analyses of included interventions revealed an unexpected direction of effect associated with the inclusion of many of the behaviour change techniques and several modes of delivery. This analysis supports research suggesting that a greater number of BCTs included in interventions does not necessarily lead to better outcomes (Dombrowski et al., 2012). This finding is in contrast with a recent review of online interventions targeting a range of health behaviours (Webb et al., 2010). Interventions that excluded ‘entertainment’ modes of delivery were associated with greater estimates of effect in response to treatment compared to interventions that were perceived to have incorporated these elements.

Study 2 explored the experiences, barriers (support needs) and facilitators (coping preferences) of paediatric pain management from the perspective of pre-adolescent children with chronic pain and their parents. This participative study (Study 2) found meaningful goal pursuit, emotion, pain beliefs and environmental factors such as the timing and lack of access to psychology services influence motivation to engage in active coping among pre-adolescent children with JIA. Important differences were noted that relate to the support needs and preferences prioritised by children and parents. For example, parents prioritise emotional distress as a target for treatment whereas children emphasise disablement (physical capability). School age children with chronic pain want support that does not isolate them further or involve additional demands that take them away from the

normality they strive to create. Participant accounts of interpersonal relationships (parental and peer support) suggest these relationships were extremely influential in shaping the coping response (Claar, Simons, & Logan et al., 2008; Walker et al., 2008; DuPen et al., 2016). Seven common categories of responses were separately identified by parent and child groups as important considerations for pain self-management. These are summarised as i) Being active (physical capability) (ii) Things you love doing (meaningful activities), (iii) Be with Mom (dependent coping habits), (iv) Emotion / relief from emotional distress, (v) Friends and practical help (social support / provision), (vi) Concentration (cognitive capability) and (vii) Find the right help (training). These findings provided support for that assertion that early intervention is necessary and coping behaviour may be more malleable if training is introduced before maladaptive coping strategies become entrenched (Schanberg, Lefebvre, Keefe, Kredich, & Gil, 1997; Hunfeld et al., 2002). Child and parent approaches to pain management were heavily influenced by the duration of the pain condition and the amount of time spent waiting for diagnosis or treatment. A theoretical analysis of study findings (i.e. categories of participant response) validated the *a priori* selection of Social Cognitive Theory as a guiding theoretical framework. Participant identified categories of responses were mapped to core SCT constructs including individual sense of agency, self-efficacy, outcome expectations, behavioural capability, behaviour regulation, observational learning, and social support. A participative research process approach was chosen and ultimately considered the best method to answer this research question.

In Study 2 and 3 we explored participant perceptions of remote therapy delivery, potential barriers and facilitators of uptake and their perceptions of successive iterations of the online *Feeling Better* intervention prototype. Participants reported remote delivery of pain self-management training would be welcomed, specifically at the outset or as soon as non-coping occurs. Qualitative findings from Study 2 revealed that a remotely delivered pain self-management intervention would be acceptable to

younger children if it helped to relieve the burden of chronic pain in a way that is consistent with how the child participant defines his or her quality of life.

In Study 3 we conducted a preliminary test of the prototype programme in terms of usability and intervention acceptability (RQ3). Mixed-methods usability testing informed four iterations of the online *Feeling Better* programme. Qualitative research was incorporated to explore intervention acceptability, barriers and facilitators of uptake of an online pain management programme from the participant's perspective. Quantitative online survey was used to assess intervention usability in terms of functionality, relevance, ease of understanding and acceptability. Medium to high website satisfaction and acceptability data suggested the prototype *Feeling Better* programme was considered usable, relevant and acceptable.

The combination of methods in this usability study were considered essential for several reasons. Firstly, it allowed an evaluation of intervention functionality using quantitative and qualitative data as intended in the feasibility trial design. This was important given the *Feeling Better* intervention was delivered using a purpose-built website and was reliant on recruitment from the community. Second, this study was an experiment in recruitment strategy. Online recruitment was quickly identified as an essential approach which allowed access to parents of children with chronic pain. Of the social media platforms used, Facebook rather than Twitter and Instagram provided the greater response. Qualitative and quantitative feedback from potential end-users about programme intensity and reading level informed important copy-editing and informed changes to the website layout and assessment which would not have otherwise been made at this late stage of intervention development. Pre and post intervention assessment measures were condensed to avoid over-assessment and the navigational choices available to participants were modified to include compulsory and free-choice features.

In Study 4 we examined the feasibility and potential effectiveness of the online version of the *Feeling Better* pain management programme (RQ4)

compared to a waitlist control in a two- arm feasibility trial. The primary aim of the study was to examine the feasibility of the project in terms of recruitment, assessment, adherence and acceptability. The findings from this feasibility study suggest that the *Feeling Better* trial functioned efficiently in terms of adherence and acceptability. However, this efficiency does not apply to the recruitment or assessment process. Recruitment was found to be time-consuming and labour intensive. Qualitative feedback at post-treatment indicated assessment fatigue may have been an issue for some participants. Future trials should rely on a combination of recruitment approaches including the addition of clinic-based, face to face recruitment and collaboration with secondary care services. Also, further consideration should be given to the battery of tests appropriate to the developmental level of participants and mode of delivery. The acceptability of technology-mediated therapy delivery for this age group was high among children and parents. Operational aspects of the intervention require further testing.

A secondary aim of Study 4 was to evaluate the potential efficacy of the online version of the *Feeling Better* programme. We hypothesised that children allocated to the Internet treatment group would show greater improvement in a range of clinical outcomes at post-treatment compared to those in the control group. The findings from Study 4 support the potential effectiveness of the online *Feeling Better* programme. The overall goals of the programme i.e. increased self-reported pain self-efficacy and use of active coping skills among pre-adolescent children with chronic pain were achieved. The *Feeling Better* programme was associated with improvement in post-treatment measures of clinical outcomes including pain intensity, self-efficacy for coping despite pain, overall quality of life, use of coping strategies and pain catastrophising. Overall, this result indicates the adaptation of the original *Feeling Better* programme and its implementation in an online platform was successful and an acceptable means of psychological treatment delivery for this age specific group and their caregivers. Treatment acceptability was high among children and parents. This may be attributed to the iterative, person-based approach used in

intervention development. This trial was under-powered as an effectiveness study; however, it did provide estimates of effect and informed methodological (operational) considerations relevant to a full-scale RCT.

7.3 Contribution of this research

This thesis contributed to a relatively neglected area of research relating to how pre-adolescent children with chronic pain respond to technology-based modes of treatment delivery. The findings of the systematic review (Study 1) provide preliminary evidence to support the effectiveness of ICT-based psychological therapies for children with chronic headache at post-treatment. Chronic pain literature is limited by a lack of focus on the intervention components that may be associated with estimates of beneficial effect. This knowledge would usefully inform the development of chronic pain interventions (Fisher et al., 2015). The findings from the intervention content analysis make an incremental advance on the literature by generating greater understanding of the potential mechanisms of behaviour change employed in existing, efficacious interventions (Study 1). No previous research has investigated the effectiveness of ICT-based, psychological interventions for paediatric chronic pain management in terms of the core components that may be associated with estimates of effect. Characterising the core components of existing interventions could usefully inform the development of future treatment studies in terms of the selection of an appropriate theoretical framework, cognitive behavioural strategies, behaviour change techniques and modes of delivery. In so doing we may facilitate replication, inform the development of effective interventions and prevent research waste.

Paediatric chronic pain literature is limited by a lack of understanding about the contextual factors that contribute to the adoption of coping behaviour in pre-adolescent children with chronic pain. Study 2 attempted to address this gap in the literature using a novel methodology designed to encourage an open dialogue on the topic (RQ2). The findings

of the participative study (Study 2) advance knowledge and generate greater understanding of the factors that influence chronic pain management among pre-adolescent children and their parents. An understanding of the lived experience of paediatric pain management in a pre-adolescent pain population is essential in order to develop effective interventions for this target group. The PRP approach has never been used to explore the lived experience of paediatric pain management.

Study 3 is the first assessment of the research and development process. This study found preliminary evidence of intervention acceptability, relevance and functionality. Study findings support the value of usability testing and of person-centered approaches to intervention development. This study demonstrated that school age children can provide end-user feedback that is valuable and practically applicable (Study 2 and Study 3). This research contributed to an iteratively developed list of core intervention components most likely to engage younger children with chronic pain in an online intervention. Importantly, no previous studies have explored the strategies and design features preferred by younger children with chronic pain or the acceptability of technology-mediated modes of therapy delivery in this cohort. This is important for researchers to understand. Exploring the mode of therapy delivery in terms of what works and for whom is important, as these intervention elements may have a modifying effect on treatment response (Ritterband et al., 2009).

The findings of Study 2 and 3 demonstrated how novel participative methodologies can be used to understand paediatric pain management and the acceptability of novel treatment strategies. Participants reported feeling empowered by the tasks set before them i.e. data generation, analysis and interpretation. The PRP approach complemented the overall aim of this research, to engage school age children in a discussion about participation in pain self-management and gave young children with chronic pain a voice in the development of a relevant treatment approach.

The findings of the feasibility trial established the previously unknown, feasibility and potential effectiveness of the adapted *Feeling Better* programme for a novel (pre-adolescent) chronic pain population (Study 4). This is valuable information because it bridges a gap between paediatric and adult focused pain management literature relating to how pre-adolescent children respond to technology-based psychological therapy. Study 4 demonstrated that online interventions can be as feasible, acceptable and potentially effective for improving perceived competence, functioning and quality of life in a younger pain population. This feasibility trial (Study 4) was the first online intervention designed specifically to support pain management for pre-adolescent children with chronic pain and their caregivers. No previous research has evaluated the feasibility of Internet-mediated, psychological therapy as a standalone source of support for this sub-group.

7.3.1 Transparent and systematic account

Several theoretical and methodological shortcomings associated with paediatric pain research have been identified. Specifically, poor report of intervention development and inconsistent application of appropriate theory and use of recommended outcomes (Craig et al., 2008; McGrath et al., 2008). This research attempted to address these limitations using an explicit intervention mapping and person-based approach consistent with best-practice guidelines for intervention development, guided by an underlying theoretical framework with consideration of developmental factors. This research demonstrated a step by step synthesis of the empirical evidence, psychological theory, expert and end-user feedback to inform intervention development. A scoping review of the literature suggests this is the first application of an intervention mapping approach to the development of a pain management intervention for children with chronic pain. In addition to intervention mapping, this is the first approach to intervention development for paediatric chronic pain management that used

the BCT taxonomy (v1) and the Mode of Delivery classification scheme (Webb, et al., 2010) to code a CBT-based intervention.

7.3.2 Application of theory

A significant contribution of this research is that it attempted to advance understanding of the cognitive behavioural approach. This was achieved by investigating the utility of Social Cognitive Theory within the context of paediatric chronic pain management. This is an important contribution as one of the main criticisms of behaviour change interventions is their lack of an explicitly reported, underlying theoretical framework or hypothesised mechanisms of behaviour change. The importance of a theoretical approach to intervention design is increasingly stressed in behaviour change research (French et al., 2012; Hawe, 2015; Michie et al., 2015). This is the first study to evaluate the utility of SCT as a means of understanding paediatric chronic pain. This research interprets study findings using social cognitive theory to understand and illustrate the experience of chronic pain and subsequent adjustment. This is an important contribution, as research in this field is novel, dynamic and replete with treatment studies which make little reference to a theoretical basis or detailed intervention content.

7.4 Relevance of findings – Intervention development

7.4.1 Complexity and intensity

The complex nature of the *Feeling Better* intervention is demonstrated by the large number of behaviours participants are asked to perform, the number of behaviours targeted within the intervention and the number of outcomes of interest. Thus, the online *Feeling Better* intervention can be called a complex intervention. While this enhances external validity and relevance in a real-world setting, it does so at the expense of internal validity i.e. a simplified context that allows control for potential confounding variables. Given the exploratory nature of the trial and

considering the multiple and competing demands of the real-world setting, including primary and secondary care, it was decided this approach would have greater relevance (French et al., 2014). Future research might consider further adaptation of the Feeling Better intervention to target specific behaviours in well-defined treatment groups.

The complexity of an intervention is separate to its intensity which refers to the commitment required of the user. Due to a lack of research focused on this area and limited guidance on developmentally appropriate, dose-response relationships, optimal intervention intensity was difficult to judge. Existing literature was consulted when deciding intervention intensity and similar interventions were used as a frame of reference (Connelly et al., 2006; Hicks et al., 2006; Palermo et al., 2009; Vigerland et al., 2016). However, parent participant feedback (Study 4) suggests that for many of the children in this age group, the intensity of the intervention was too much. This is despite efforts to reduce programme intensity during the development phase (Chapter 4 and 5). On the other hand, the number of pain behaviours targeted in the *Feeling Better* intervention was viewed very favourably by parents. It is possible that programme intensity would be diminished by spreading the clinical content of the programme further and extending the programme from 9 weeks to 12 weeks in duration and restricting the time commitment required to complete a treatment session. For example, March et al (2009) conducted an online CBT-based intervention for children with anxiety disorders. The programme (BRAVE online) was rolled out over a ten-week period for children aged 7-12 years. Response to treatment was favourable for the Internet intervention group and of note here is the duration of treatment specific to the pre-adolescent target group (March, Spence, & Donovan, 2009). Questions remain regarding the optimal dose of treatment, the maintenance of effects over time, the influence of the online tutor in supported versus unsupported interventions and the effectiveness of ICT-based psychological treatments other than CBT.

7.4.2 Difficulties associated with the application of guidelines

Careful consideration should be given to the balance between top-down (theory-driven) and bottom-up (person-centered) approaches to intervention development when selecting guiding intervention development frameworks. There were many challenges associated with the current attempt to integrate evidence and information from different sources i.e. patient feedback on support needs and preferences, the context in which pain management is undertaken and evidence-based guidelines for pain management. This research undertook a systematic and transparent approach to intervention development that offers the potential for replication, reduction of research waste and valuable information that contributing to our understanding of the potential mechanisms of action (French et al., 2012). However, it also skewed the focus of intervention development toward a top-down (theory-driven) rather than balanced intervention development approach. Challenges emerged when attempting to implement selected CBT strategies, BCTs and modes of delivery in a way that served their parameters for effectiveness, aligns with the guiding theoretical framework and best practice guidelines and meets the support needs identified by end-users. The criteria for one often conflicted with another. For example, the implementation of certain BCT's in a manner consistent with their definition and parameters for use was impeded by conflicting demands of adhering to the principles of clear and effective communication for the target group. To adhere to both the rules of communication (e.g. one idea per session page, one-line sentences, simple language, active voice) and the necessarily strict definitions of BCTs would have resulted in a much larger, unmanageable and over-intensive intervention for this pain population. The decision was made to strike a balance between form and function. To both simplify the form and attempt to retain the function of the technique. This resulted in many of the incorporated BCTs being coded as likely present rather than definitively.

Upon reflection, understanding of the challenges of paediatric chronic pain management might have been better demonstrated by adherence to a bottom up approach to intervention development. For

example, the findings from Study 2 suggest the optimal intervention for pain management is one that is simple, easy to use and understand and does not isolate the user further (i.e. may be incorporated into day to day routines). The obligation to meet all guidelines for best practice meant the final intervention was too complex and intense for the target group. The difficulties associated with the implementation of the included intervention development guidelines are discussed below.

7.4.3 Intervention mapping

This research demonstrated an application of the intervention mapping protocol with a relatively high degree of specificity in the context of a paediatric chronic pain. The 2008 MRC guidelines and intervention mapping protocol were instrumental in developing and maintaining clarity of purpose in terms of what this research was trying to achieve. The intervention mapping protocol was considered a useful but cumbersome and intensive. The advantages of IM are that it provides an incremental, practical structure to guide the intervention development process. This facilitated the systematic use of theory, empirical evidence and end-user perspectives in intervention development. Needs assessment was helpful to understand the chronic pain problem and specifically gaps in literature and practice which might be addressed. This process helped to identify programme goals and essential components for the *Feeling Better* intervention which are based on a detailed assessment of the evidence base and participant needs. The creation of matrices of performance and change objectives facilitated an explicit account of the development process potentially allowing for easier replication of the intervention. Intervention programme planning in Step 4 allows the ongoing refinement of the prototype programme in response to the findings from each work-package. Early consideration of adoption and implementation issues may have contributed to favourable recruitment and adherence outcomes. Planning for a full evaluation of the intervention identified issues with outcomes and measures which would not have been highlighted otherwise such as over-

assessment and cumbersome informed consent procedures (Dansky, Thompson, & Sanner, 2006).

The disadvantages of the IM protocol are that the process of creating the behaviour change matrix for a complex, multicomponent intervention such as this is extensive and time consuming (Michie et al., 2008; Hurley et al., 2016; Kok et al., 2016). The specificity of the IM protocol was difficult follow within the context and constraints of this research. Access issues were such that it was decided early in the development process to focus recruitment efforts on the primary stakeholders, pre-adolescent children with chronic pain and their care-givers. Recruitment challenges and time constraints prevented the involvement of health professionals currently working in a healthcare setting in the Republic of Ireland. This meant a very homogenous group contributed to needs assessment and user-testing. In line with Hurley and colleagues (2016) this study veered from the typical IM recommendation in that the decision to use an online mode of treatment delivery did not evolve organically in Step 4 but was rather confirmed by needs assessment prior to that stage. The research team had a good idea from the outset that there was a need for a widely accessible and developmentally appropriate pain management programme. The team also had access to clinical content in the form an established CBT-based pain management manual for carers of individuals with chronic pain and intellectual disability (McManus & McGuire, 2010). The decision to adapt an evidence-based, manualised programme comprising high quality materials and clinical content allowed the development process to focus on the practicalities of adaptation and implementation. For example, copy-editing the clinical content and materials to the appropriate level of development, engaging training for the online tutor and translation to an online platform.

7.4.4 Retrospective coding

Issue with retrospective coding include inaccurate representation of intervention content when coding is based on inference from text. This is a significant issue in this field of research, in which very few studies report

the explicit use of theory or the content in both the intervention and control group. Retrospective coding does not capture the fidelity (i.e. uniformity of intervention delivery) or account for suboptimal performance of therapeutic techniques (Hankonen, 2014). For example, the findings of Study 1 and Study 4 are in contrast with those of a systematic review conducted by Webb et al., (2010) which suggests interventions with a theoretical basis and interventions that incorporate more BCT's are more effective (Webb, et al., 2010). This discrepancy may be attributed to inaccuracy in the coding process, poor report of intervention content, the use of content analysis frameworks that are lacking (BCTT, TDF) or simply that these components did not achieve their function due to poor implementation or lack of relevance (Chapter 3).

7.4.5 Behaviour change techniques

In contrast with Webb et al., (2010), both the systematic review (Study 1) and the feasibility trials (Study 4) found that the number of BCTs considered present in the intervention was not directly associated with estimates of beneficial effect. This finding is in line with meta-regression analyses that have noted that increasing the number of BCTs in an intervention is not necessarily associated with better outcomes (Dombrowski et al., 2012). The purpose of the Behaviour Change Technique Taxonomy, Version 1 (BCTT) (Susan Michie et al., 2013) is to provide intervention developers with a universal language with which to describe the active ingredients in interventions and to promote transparency in intervention design thereby facilitating easier replication (Michie et al., 2013). This research found the BCT taxonomy v1 to be a useful but inflexible tool with which to characterise the *Feeling Better* intervention content. This research agrees with previous criticisms of the BCTT as an overly systematic and inflexible approach to understanding behaviour change, theory and practice (Ogden, 2016). The BCTT v1 fails to account for the influence of interpersonal factors on behaviour change and is limited by a lack of consideration of developmental factors. It also fails to account

for the frequency and intensity with which BCTs are presented. This is an important consideration as different frequencies could have a different impact on intervention outcomes. Finally, the lack of clarity surrounding the role of the website platform as a treatment delivery system should be addressed in future research

Despite reference to the relevant literature and training, there was some confusion as to how the code BCTs in remotely delivered interventions. For example, is the *Feeling Better* website a means of treatment delivery that could change the nature of treatment or is it a mode of delivery i.e. akin to a syringe or patch (Eccleston, 2011)? This research was guided by Ritterband's model of internet intervention development which suggests that change in outcome will be mediated by the clinical content of the intervention and use of the intervention (Ritterband et al., 2009). Some BCTs are defined in such a way that they accommodate Internet-mediated interventions e.g. BCTs 10.4 social reward, defined as verbal or non-verbal reward. Whereas others are more difficult to code e.g. the BCT 15.1 verbal persuasion about capability or the BCTs that refer to positive reinforcement. In the online platform, these BCTs were implemented using automated messages which were periodically delivered and, in some cases, based on accomplishment i.e. task completion. In addition, support from the online tutor was delivered via internal email or instant messaging. These and other BCTs were coded as possibly present because their functions were carried out by the operating system rather than delivered verbally or by a therapist. In summary the BCTT was considered a useful but inflexible framework for retrospective (Study I - Chapter 3) and prospective evaluation of online intervention content (Study IV- Chapter 6). The BCT Taxonomy was also found to be inflexible in terms of developmental perspective and the accommodations that were necessary to apply BCTs to younger age groups. It may be that the definition of certain BCTs is too restrictive.

7.5 Relevance of findings – End-user

7.5.1 Parental role

As expected, level of development seemed to influence the nature and involvement of parents in treatment sessions and in the correspondence between sessions. Correspondence was more frequent with parents of younger children in the target age range. These parents asked more questions of the online tutor than parents of older children, or child participants in general. Further research is required to determine the optimal level of parental involvement for this age group and treatment setting. Improvement in parental protectiveness at post-treatment and qualitative feedback would suggest this level of involvement and an information-only section is enough to motivate change. However, tailored parent training, has been associated with improved child anxiety and motivation to engage in treatment and family functioning (Thienemann et al., 2006; Lebowitz, Omer, Hermes, & Scahill, 2014; Palermo et al., 2016). Further research is required to determine if parents can be effectively engaged as lay-therapists.

7.5.2 Meaningful involvement of end-users

A common criticism of qualitative research in relation to intervention development is that the former does not always meaningfully influence the latter (Yardley et al., 2015). Study 3 and 4 demonstrated that it is possible to combine empirical evidence with theory and end-user feedback in a meaningful way that significantly contributes to intervention usability and acceptability. Mixed methods, user-testing (Study 3) highlighted key features, content and design principles preferred by this population. Research findings (Study 2 and 4) indicated a need for cognitive strategies for behaviour change. However, the preferred coping strategies reported by children in Study 2 were behavioural (Thastum et al., 2005). It may be that children with different pain conditions would more readily incorporate cognitive strategies for coping. Previous research suggests this preference is also associated with earlier levels of development indicating this result was inevitable (Kingery et al., 2006).

Feedback from this iterative study informed the combination of components and design principles which ultimately comprised the final intervention. Collaboration with end-users and experts in each phase of intervention development is believed to have directly contributed to the acceptability of the *Feeling Better* intervention. A focus on usability and acceptability prior to the feasibility trial was justified on the basis that behaviour change may be mediated both by intervention content and by use of the intervention itself (Ritterband et al., 2009). Moreover, the exploratory nature of this trial, the use of a purpose-built website and the recruitment approach (i.e. from the community) necessitate comprehensive usability testing to anticipate functionality or other usability issues that might have a negative impact on the user's experience. This research shows that qualitative feedback can translate to meaningful change in the topic of interest.

7.5.3 User engagement

In depth, mixed methods research is recommended to better understand how individuals engage with ICT-based interventions (Yardley et al., 2016). Engagement may refer to the users' relationship with technology or to the users' engagement with the therapeutic process or both (Ritterband et al., 2009). The former may be measured in terms of objective user access data and the latter through user attempts to achieve positive cognitive, emotional and behavioural change. This research identified a number of factors that may have influenced engagement with the *Feeling Better* intervention. The testing process was multi-faceted therefore it is difficult to discern which elements were associated with user engagement in the feasibility trial. However, the combined, objective usage data and subjective treatment satisfaction data suggest goal setting, progress review and self-monitoring were among the most important techniques promoting engagement in the online intervention. Participant feedback (Study 3 and 4) suggests progress review (i.e. personalised correspondence relaying progress toward goals, positive reinforcement, reminders to practice,

complete homework and return to the next treatment session) was particularly influential in terms of adherence and treatment acceptability.

Further consideration should be given to how engagement and adherence is defined – this research assessed treatment adherence in terms of session and homework compliance. Mediation analyses might explore the effects of age and types of treatment compliance e.g. weighted compliance. Future research should focus on developing and testing objective and subjective models and measures of effective engagement (Cavanagh, 2010; Morrison et al., 2014; O’Connor et al., 2016; Short, Rebar, & Ronald, 2013; Yardley et al., 2016).

Despite the current effort to influence engagement, there remain many modifiable and non-modifiable factors that may inhibit uptake of the *Feeling Better* intervention e.g. literacy, education background, cognitive capabilities and support needs (Cashen, Dykes, & Gerber, 2004). Emerging guidelines for how this can be achieved are welcomed (Health Information & Quality Authority [HIQA], 2014).

While the study design does not allow a comparison, these findings contribute to literature exploring the value of supported internet interventions comparison with unsupported interventions (Leykin, Muñoz, Contreras, & Latham, 2014; Rheker, Andersson, & Weise, 2015). The provision of “support on demand” in addition to the Internet CBT intervention was extremely labour intensive and therefore difficult to maintain. The scalability of the *Feeling Better* intervention would certainly be improved by research that compares supported and unsupported online interventions.

7.6 Relevance of findings – Delivery and assessment

7.6.1 Website development

Positive user feedback (Study 2, 3 and 4) supports the decision to use a purpose-built website rather than use open-source software so that most

of the CBT- strategies could be delivered online without extensive deviation from how they might be presented in a face to face setting using the original manual. Additionally, the advantages of a purpose-built platform included the implementation of CBT strategies in an engaging way using puzzle/game-like features such as “spot the difference”, “drag and drop” the correct answer, complete the positive coping sentence and so on. Many of the interactive features of this programme would not have been feasible using open-source software. Moreover, according to the need’s assessment, the *Feeling Better* programme would engage more children if it was accessible across a range of portable devices e.g. tablets and this was not feasible using the available, free open source software.

Despite these advantages, there were implementation challenges related to finding the right balance between achieving the function of a given CBT strategy e.g. problem-solving skills training and the optimal presentation of the strategy in a way that will appeal to the target population. Younger children struggled to complete these exercises without assistance from parents. While it was always intended that parents would be proactive in their role of lay-therapist, it seems more work is needed to simplify these strategies for younger participants. It is likely therefore that website design by a professional might contribute to better outcomes in terms of acceptability, ease of use and understanding. This observation is supported by Study 4 usage and qualitative data which suggests further work is needed to effectively adapt the more sophisticated CBT-strategies e.g. cognitive-restructuring, problem-solving and communication strategies for this population.

7.6.2 Chronic pain assessment and outcomes

A highlight of this research is that it assessed several of the recommended core outcomes for chronic pain and can contribute to empirical evidence informing the development of a pilot intervention. Outcomes consistent with the proposed mechanisms of action were selected.

Appropriate psychometric measures of these outcomes were chosen based on the recommendations of the PedIMMPACT committee (McGrath et al., 2008). However, assessment was hampered by a lack of psychometric instruments that meet the criteria of being both recommended as a measure of a core outcome (e.g. physical functioning) and appropriate to participant age and chronicity (i.e. designed for children under eight years with chronic rather than acute pain). For example, the only age appropriate, recommended measure of physical functioning and psychological distress (anxiety and depression) was the Pediatric Quality of Life Inventory which contains relevant subscales: physical health and psychosocial health (Varni et al., 2001). This issue extended to assessment of specific outcomes. Similar issues were experienced when searching for appropriate measures of pain catastrophizing and self-efficacy, this is despite a growing body of evidence that suggests children can appraise their pain experience from five years of age (Beyer & Wells, 1989; von Baeyer, Uman, Chambers, & Gouthro, 2011). Study 4 opted to use a validated pain catastrophising scale for children and parents despite it being designed for children age eight years and older.

There were also challenges associated with the selection of appropriate psychometric instruments that are suited to therapy delivery. Advances in technology allow real-time, ecological data collection which could pose a problem for researchers attempting to select validated, evidence-based methods of assessment suited to remote delivery (Eccleston & Crombez, 2017). Innovations in ICT-mediated therapy delivery mean therapy can more easily be tailored to individual needs and problems and to rapid, real-time data collection. While the *Feeling Better* programme was not tailored to individual needs, it was tailored to the needs and potential problems of an age-specific group and pain coping experiences in the natural environment. The method of therapy delivery afforded regular opportunity to check-in with the participant i.e. check-in at the beginning of each treatment session. In turn, this allowed periodic assessment of progress on functioning (e.g. how many days of school were missed this week). In this instance, questions regarding role and physical functioning were

developed specifically for the *Feeling Better* programme that might assist assessment of ongoing progress. However, methods of assessment and evaluation will need to be developed that keep pace with innovations in therapy delivery (Eccleston & Crombez, 2017).

7.7 Relevance of findings – Theoretical basis

Social Cognitive Theory (1996; 2005) is an encompassing account of behaviour and behaviour change that seems to address many of the key determinants identified in the pain literature. It could be argued the breadth of SCT is too encompassing. The broad reach of this theory means it is difficult to operationalise in its entirety (Bandura, 2005; 2006). The only SCT construct not referenced in the current research is self-regulation of conduct. This refers to the moral justification of a behaviour by an individual who is acting in a self-serving way or aware in some way that they are engaging in a practice that is not entirely beneficial. Initial validation of the SCT considered the practice of over-protective parenting behaviour as a determinant that maps to this construct however this was discarded due to uncertainty surrounding the accuracy of this assessment and the fact that the determinant applied to parents and not children i.e. the patient group.

Moreover, the same criticism could be applied to other theories of health behaviour change including the theoretical domains framework (Cane et al., 2012). Here, the use of SCT is justified on the basis that the health problem is complex and multidimensional requiring consideration of the broad range of biopsychosocial factors that shape pain behaviour. Also, in line with SCT the mechanisms of change within CBT are considered bi-directional in effect, whereby therapeutic change in one area e.g. pain beliefs will have an influence on another e.g. pain behaviour.

The current research specifically targeted self-efficacy, outcome expectations, behaviour regulation and social support as significant predictors of pain intensity and pain interference. This is based on a wealth of research that has evidenced the role these factors play in influencing and reducing the symptoms associated with chronic pain. However, other

research has also shown that children's pain response can also be influenced by a range of factors such as pain severity, peer support, parental history of chronic pain. Thus, to enhance the applicability of the current findings future research should strive to compare the effect of self-efficacy and outcome expectations with other known predictors. Assessing the effect of these predictors simultaneously may help the development of more robust disability reduction techniques among children.

SCT explains how pain behaviour might be impacted by the CBT process to effect change in the pain experience and pain management. This process was mapped at the outset (see Appendix 2 and 3) and a theoretical model of behaviour change is illustrated below (Figure 7.1). The proposed logic model is an attempt to illustrate how the intervention might be expected to operate. The current systematic review and qualitative research identified SCT as a useful theoretical basis for the online *Feeling Better* intervention. The *Feeling Better* manual and subsequently the online clinical content is comprised of CBT techniques that were selected and implemented in the intervention website to address key determinants of pain behaviour (i.e. SCT constructs). Since the *Feeling Better* programme is based on CBT principles, it was hypothesised that a tailored pain management programme that targets participant pain beliefs and appraisals (e.g. control over pain, self-efficacy and catastrophic appraisal), would enhance self-efficacy for coping despite pain and use of coping strategies. That cognitions would be shaped by perceived barriers, social support and behavioural habits, skills and goals. Understanding these influences at a theoretical level usefully informed the *Feeling Better* intervention and may contribute to the design of effective and more engaging therapy interventions. Furthermore, it is proposed that interventions targeting these constructs are likely to modify the individual's sense of competence and coping behaviour. Adaptive change in self-efficacy and coping behaviour is likely to lead to better adjustment to chronic pain and reduced pain-related disability. Explaining the determinants of pain behaviour and adjustment in terms of SCT may help to inform future intervention development by specifying targets for treatment and highlighting potential barriers to the uptake of psychological

support.

The theoretical constructs targeted in the *Feeling Better* intervention are comparable to those identified in similar, online interventions. Retrospective intervention content analysis identified the likely theoretical basis of existing, online interventions for paediatric chronic pain management (Study 1). Retrospective analysis of the intervention content, reviewed in Chapter 3 found existing ICT-interventions for paediatric pain most frequently target “knowledge”, “skills” and “behaviour regulation” as well as “beliefs about consequences”, beliefs about capabilities” and “environmental contact and resources”. Interestingly, this content analysis indicates existing ICT-based therapies for paediatric chronic pain target “goals”, “emotion” and “reinforcement” less frequently. This is in contrast with the *Feeling Better* programme which prioritised these constructs as targets for treatment and included strategies to address each in the opening section (check-in) of each module. The decision to prioritise meaningful (self-selected) behavioural goals is based on SCT, qualitative findings from Study 2 and empirical research that suggests the child’s coping response may be mediated by meaningful goal pursuit (Fisher & Palermo, 2016).

7.7.1 Implications for research and practice

Based on the findings of this research, it is clear the evidence base is not keeping pace with current interest in the development and evaluation of ICT-based therapy interventions (Eccleston, 2011; Morley et al., 2013; Eccleston & Crombez, 2017). Research is needed that contributes to current understanding of how and for whom novel treatment strategies are expected to work. A need for higher quality trials is indicated. This should involve large sample sizes, well-defined target populations, evaluation of different therapies and higher standards of report.

Capturing a more comprehensive picture of commonly targeted theoretical domains, behaviour change techniques and modes of delivery incorporated in existing online interventions is only a first step. Further research is required to explore the optimal combination of intervention

components. Future studies might employ advanced statistical techniques to explore the synergistic and cumulative effects of different combinations of core components (e.g. therapeutic strategies, BCTs and MoDs (Webb, et al., 2010; Dombrowski et al., 2012; Morrison et al., 2012; Michie, Johnson, & Johnston, 2015; Morrison, 2015; van Genugten, Dusseldorp, Webb, et al., 2016) for specific chronic pain conditions and population groups.

School age children can provide important and valuable information relating to their own pain management and end-user feedback can be meaningfully applied in intervention development. Researchers and clinicians should aim to engage children with chronic pain as active agents in their own treatment protocol. It is important to understand the factors that influence coping behaviour in this population in order to develop effective intervention. Children may be motivated to engage in pain self-management using meaningful goal pursuit, guided treatment delivery and tailored clinical content. Evaluation of these factors may help explain individual differences in response to treatment or coping style. Future research may also consider tailoring the clinical content for specific pain conditions and thereby contribute to current understanding of what works and for whom. Interventions that target self-efficacy beliefs, outcome expectations, behaviour regulation and social support could enhance individual sense of personal agency and capacity for coping with chronic pain.

Social cognitive Theory may be a useful framework with which to understand how the *Feeling Better* and similarly dedicated intervention might work. It is important to develop a theoretical understanding of the variation in the utility of the CBT treatment approach under certain circumstance. Preliminary support for the use of SCT is also an indication of support for the way the theoretical mechanisms were measured. Therefore, future research may benefit from testing the effect of these moderators in more rigorous experimental designs or the way in which the intervention was conducted. Further research is necessary to determine if this framework is useful only for this intervention or may be used as a

framework for interventions focused on other chronic conditions.

Healthcare professionals were unwilling to get involved in an exploratory trial (Study 4). It is expected that as the quality of the evidence base and research improves, healthcare professionals may become less reticent about collaboration in studies using novel modes of treatment delivery. Due to the inefficiency of the current recruitment approach, collaboration with secondary care centres should be incorporated with recruitment from the community in future trials. However, despite the inefficiency of the current approach, this research suggests social media can be a useful tool in the recruitment process. Most participants were recruited via social media and online parent-led, support networks. Although small, the sample size in each of these studies was sufficient to answer the research question.

In Study 4, the conversion rate from enquiry to study enrolment was high and the randomisation process achieved equality between groups which were relatively equal in size and well balanced. This supports the decision to streamline and embed the recruitment, registration (information and consent process) baseline assessment and randomisation procedures. This observation may usefully inform the design of a larger trial.

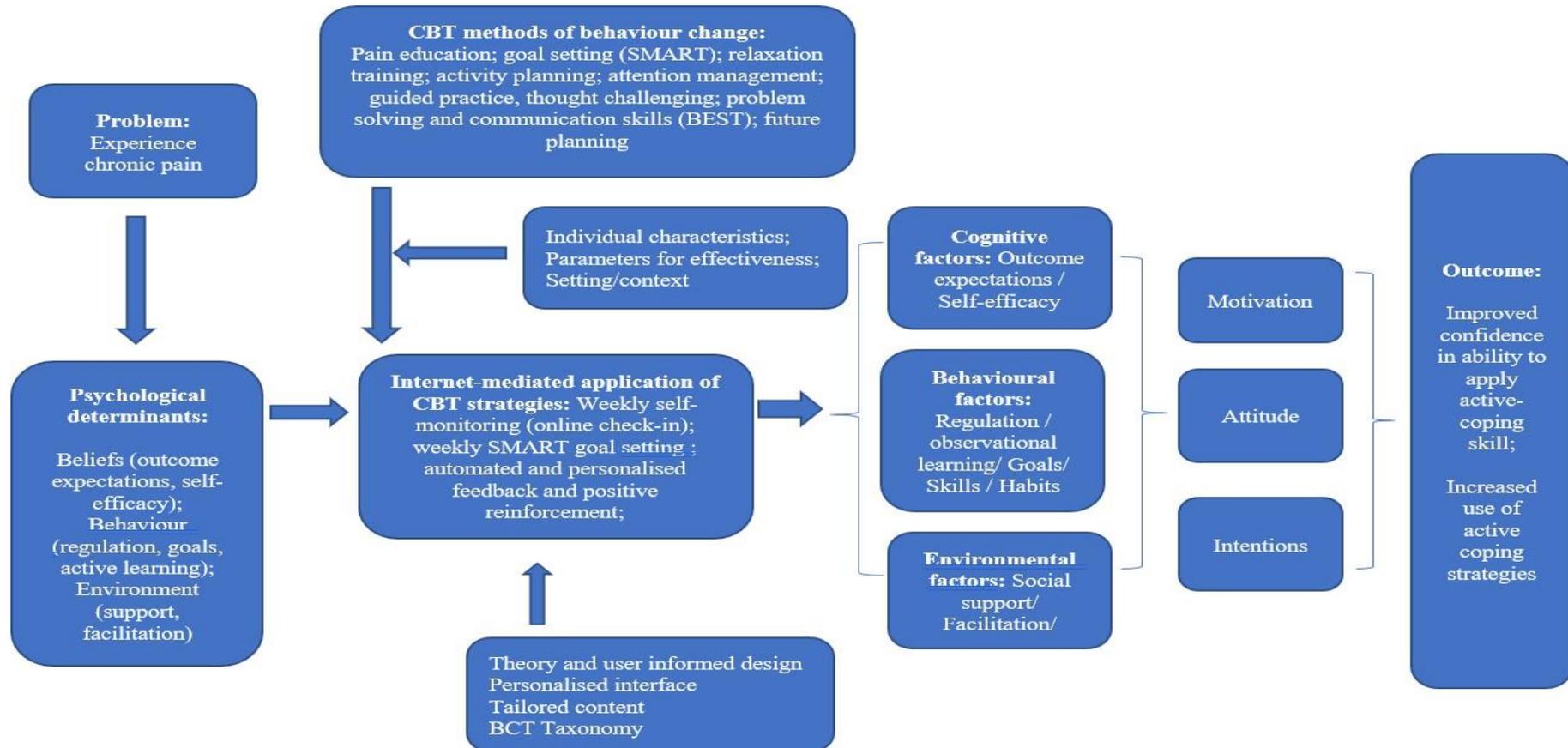
Loss to follow-up and participant feedback in Study 4 suggest assessment fatigue may have contributed to loss to follow-up observed at Time 3 (three months). Despite four iterations of intervention development and refinement further testing is recommended. In Study 4, the acceptability of the *Feeling Better* programme was high despite the identification of several areas for improvement (e.g. optimal intensity and treatment-dose for this age-specific cohort, improved readability and usability and a revision of assessment procedures to prevent assessment fatigue). These limitations should inform the design of a larger trial.

The findings from the *Feeling Better* trial suggest the potential for beneficial effect in child reported pain self-efficacy, pain catastrophizing, overall quality of life and use of coping skills. These effects may be treatment specific (i.e. indicative of absolute efficacy) or due to non-specific

effects of treatment (i.e. placebo). The exploratory nature of the *Feeling Better* trial prevents further conclusion however these effects are promising and support the assertion that a larger pilot trial is warranted.

Overall, this research suggests that ICT-based therapy delivery may provide a novel, viable solution to resource and access issues that prevent uptake of and access to psychological support for paediatric pain management. Important targets for treatment were identified in Study 1 and 2, that if addressed may contribute to the effectiveness of interventions designed for paediatric pain management

Finally, significant improvements are possible for this target population. In Study 4, pre-adolescent children with chronic pain showed significant improvement in several key clinical variables following treatment that was delivered over the Internet in comparison with a waitlist control group. Also, providing parents with the information and materials necessary to facilitate child pain self- management has the potential to (i) interrupt maladaptive parenting behaviour (i.e. habits) and (ii) encourage the child's use of adaptive coping strategies.



7.7.2 Figure 7.1 Hypothesised Logic Model for the *Feeling Better* intervention

7.7.3 Strengths and limitations

This research has a number of strengths and limitations which are outlined below:

In Study 1, a systematic review, meta-analysis and intervention content analysis of psychological therapies delivered using information and communication for paediatric chronic pain management was conducted. The strengths of this review include the use of a pre-established and published review protocol (Traynor et al., 2016); the use of a search strategy that included published and unpublished RCTs; assessment of recommended core outcomes (McGrath et al., 2008) and study risk of bias and quality assessment. This research was limited by several methodological issues. The search strategy was unsuccessful in attempting to extract unpublished trials from grey literature. Only ten trials were included (N=676 participants) which limited the conclusions that can be drawn from meta-analyses. The quality of the included trials was low to moderate, reduced by a lack of detailed trial reporting, small sample sizes and a reliance on passive control conditions. Finally, some meta-analyses of primary and secondary outcomes were prevented due lack of data. Therefore, meta-analysis was supported by a narrative synthesis and intervention content analysis. The findings from this review identified gaps in the literature and provides an overview of the evidence base.

In Study 2, a participative qualitative study was conducted which explored the lived experience of chronic pain management from the perspective of pre-adolescent children with chronic pain. The strengths of this study include the use of a developmentally appropriate and empowering participative protocol and the involvement of participants in each phase of research (i.e. data generation, analysis and interpretation). The process of creating schematic maps helped to reduce the cognitive demands placed on child participants and was a fun, creative and engaging way to explore coping for the child's perspective (Nelson & Tusaie, 2011). As such, this methodology may have elicited more information that would otherwise

have been captured.

The limitations of this study include the likelihood of bias associated with the presence and profile (age, status, gender) of the researcher. While efforts were made to ensure the comfort of participants, the university setting for two of the participative workshops is also likely to have led to bias. Another limitation of this research refers to the homogeneity of the sample. A more heterogeneous group including child participants with other chronic pain conditions and the inclusion of other healthcare professionals might have offered a more varied perspective on the daily challenges of paediatric pain management. The findings from this study informed current understanding of the factors that influence the adoption of coping behaviour in a relatively under-researched and under-served target pain population.

In Study 3, a mixed-method usability study was conducted which combined a participative protocol and online user-testing. This study was designed to test study usability and acceptability prior to a feasibility trial. The strengths of this study include the use of multiple and diverse methods of user testing and the involvement of expert and end-user feedback in intervention development. The limitations of this study include the use of staggered user-testing of treatment modules as opposed to complete programme evaluation which might have identified issues relating to programme intensity and complexity. The findings from this study informed 4 iterations of intervention development and shaped the development of a person-based intervention for paediatric pain management.

In Study 4, a two-arm, parallel feasibility trial was conducted to evaluate the feasibility and potential effectiveness of an online version of the Feeling better pain management programme for pre-adolescent children with chronic pain and their care-givers. The strengths of this study include the efforts made to identify the characteristics of completers compared to non-completers. Important differences were identified which suggest participants have a higher risk of drop out and may require more support if they report higher and inferior scores on measures of psychosocial health,

pain catastrophising and treatment expectations at baseline. This study has several limitations. For example, this study uses a simple trial design which limits the conclusions that may be drawn. This research is also largely reliant on self-report methods, which depend on the child's ability to access their feelings and on their willingness to report them. Such measures are susceptible to response biases (e.g., self- presentation), particularly on socially or personally sensitive topics.

7.8 Conclusions

This programme of research has demonstrated that the adapted *Feeling Better* programme has the potential to make an important contribution within the field of Internet- mediated treatment delivery for paediatric pain. The four, inter-linking studies that comprise this thesis, collectively support the potential of (i) remote therapy delivery as a viable solution to access and resource issues which prevent uptake of or access to psychological support for chronic pain management and (ii) the *Feeling Better* programme as an effective source of support for chronic pain management among pre-adolescent children and their care-givers.

The online *Feeling Better* programme was developed in line with best practice guidelines for the application of evidence, theory and end-user perspectives (Michie et al., 2008; Michie & Johnston, 2012). This is the first study of this nature that attempted to promote pain self-management among pre-adolescent children with chronic based on the core components of SCT and mediated by online treatment delivery. The *Feeling Better* programme is an innovative, interactive, user-friendly and engaging approach to teaching pain self-management skills for younger children with chronic pain and related symptoms.

This research makes a novel contribution to the paediatric pain management literature using Internet-mediated psychological interventions. The current intervention advances research on evidence-based CBT, offering new insight into content and context critical for treatment delivery among younger pain populations. This method of treatment may have

potential as a source of supplementary support for families coping with paediatric chronic pain and may be provided to those on waiting lists, as an adjunct to current face to face psychological therapy or as a supplement to standard medical care for paediatric chronic pain management.

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Appendices

Appendix 1 Research Ethics Committee Approval Letters from NUI Galway



Leas-Uachtarán
um Thaighde

Vice President
for Research

Date: 8th January 2014

Ref: 13/NOV/01

Angeline Traynor
PhD Candidate Psychology and Health
Arts Millennium Building Extension (AMBE)
School of Psychology
National University of Ireland
Galway

Dear Ms Traynor

Re. Ethics Application:
Feasibility and effectiveness of an online version of the *Feeling Better* pain management programme for young children and their care-givers

I write to you regarding the above proposal which was submitted for Ethical review. Having reviewed your response to my letter, I am pleased to inform you that your proposal has been granted **APPROVAL**.

All NUI Galway Research Ethic Committee approval is given subject to the Principal Investigator submitting annual and final statements of compliance. The first statement is due on or before 31st October 2014. Please see section 7 of the REC's Standard Operating Procedures for further details which also includes other instances where you are required to report to the REC.

Yours Sincerely

Allyn Fives
Chair, Research Ethics Committee

Appendices



OE Gaillimh

NUI Galway

Leas-Uachtarán

um Thaighle

Vice President

for Research

Date: 7TH April 2016

Ref: 13/ NOV/ 01

Angeline Traynor

PhD Candidate Psychology and Health

Arts: Millennium Building Extension (AMBE)

School of Psychology

National University of Ireland

Galway

Dear Ms Traynor

Re. Amendment of Ethics Application 13/ NOV/ 01: Feasibility and effectiveness of an online version of the *Feeling Better* parental management programme for young children and their care-givers

I have read your proposed amendment to the above mentioned study. On behalf of the committee I am happy for the amendments to be given FULL APPROVAL.

Yours Sincerely

Allyn Fives

Chair, Research Ethics Committee

Appendix 2 Intervention Matrix

Intervention map linking change objectives to methods and practical applications

SCT / evidence-based determinants and change objectives of pain self-management	Performance objectives	Module No.	Methods / Strategies	Behaviour change techniques as per BCT taxonomy v1 (Michie et al. 2013)	Mode of delivery Scheme (Webb et al, 2010) *
Desired outcome 1: enhance participants self-efficacy by end of programme and 3-month follow-up					
<p>Knowledge</p> <p>develops an understanding of chronic pain / types of coping / chronic pain cycle; increase motivation to engage in ACB.</p>	PO.1, PO.6	1-8	Information; modelling; persuasion; personalising risks and benefits;	5.1., 5.3., 5.6.,	a, b, c, e, f, g, i, j, o, p, r, s, t
<p>Self-efficacy</p> <p>builds confidence in ability to use CBT strategies; develops a sense of agency; encourages adaptive beliefs about capability;</p>	PO.5, PO.8,	1-9	Information; modelling; persuasion; personalising risks and benefits; barrier / facilitator identification; modelling; personalising risk; reinforcement; implementation intentions	1.1, 1.2.,1.4., 2.4., 3.1., 4.1., 4.2., 5.1., 6.1., 8.1., 8.2., 8.3., 15.1, 16.2.	a, b, c, e, f, g, i, j, o, p, r, s, t

Skills						
describes the importance of practicing SM skills; Develops an understanding of how and when to use CBT strategies	PO.5, PO.6, PO.7, PO.8	1-9	Relaxation training; action planning; attention management skills; communication skills; thought challenging; problem-solving skills; communication skills; future planning	1.1.,1.2., 1.4., 1.6., 4.1., 4.2., 5.1., 6.1., 8.1.,8.2., 8.3., 11.2.,	a, b, c, e, f, g, i, j, o, p, r, s, t	
Self-regulation						
increases motivation / intention to monitor progress in pain SM	PO.2, PO.4, PO.5, PO.6, PO.7	2-8	Information; modelling; persuasion; personalising risks and benefits; modelling; graded task setting;/action planning; implementation intentions; barrier / facilitator identification	1.1, 1.4., 1.6., 2.4., 3.1., 4.1., 4.2., 5.1., 6.1. 8.1. 8.2, 8.3., 11.2.	a, b, c, e, f, g, i, j, o, p, r, s, t	
Behavioural goals						
increase motivation / intention to set and pursue goals; develops an understanding of SMART goals and how they relate to pain SM	PO.2, PO.4, PO.5, PO.7	1-8	Information; modelling; action planning; implementation intentions; SMART goal setting; barrier / facilitator identification	1.1, 1.4., 1.6., 2.4., 3.1., 4.1., 4.2., 5.1., 6.1. 8.1. 8.2, 8.3.,	a, b, c, e, f, g, i, j, o, p, r, s, t	

Behavioural capability improves ability to recollect what they have learned	PO.6	1-8	Information; modelling; action planning; implementation intentions; SMART goal setting; barrier / facilitator identification	1.1, 1.4., 1.6., 2.4., 3.1., 4.1., 4.2., 5.1., 6.1. 8.1. 8.2, 8.3.,	a, b, c, e, f, g, i, j, o, p, r, s, t
Social support develops an awareness of social support	PO.5,	1-9	Information; modelling; action planning; implementation intentions; barrier / facilitator identification	1.1, 1.4., 2.4., 3.1., 4.1., 4.2., 5.1., 6.1. 8.1. 8.2, 8.3., 11.2.,	a, b, c, e, f, g, i, j, o, p, r, s, t
Facilitation develops an awareness of available resources; typical barriers and facilitators of pain SM	PO.3, PO.6,	2-8	Information; relaxation information /training; barrier / facilitator identification	1.1, 1.4., 1.6., 2.4., 3.1., 4.1., 4.2., 5.1., 8.2, 8.3.,	a, b, c, e, f, g, i, j, o, p, r, s, t

Desired outcome 2: increase use of CBT-based self-management strategies by participants by end of programme and 3-month follow-up					
Knowledge					
gain insight into effective/ineffective coping styles through practice	PO.9	1-8	Information; SMART goal setting; graded tasks; modelling; persuasion; personalising risks and benefits; barrier / facilitator identification; modelling; active participation.	5.1., 5.3., 5.6.,	a, b, c, e, f, g, i, j, o, p, r, s, t
Self-efficacy					
increase self-efficacy for functioning despite pain using CBT strategies	PO.10, PO.11, PO.13	1-9	Information; self-regulation; SMART goal setting; graded tasks; actioning planning; modelling; persuasion; personalising risks and benefits; barrier / facilitator identification; modelling; personalising risk; implementation intentions	1.1, 1.2.,1.4., 2.4., 3.1., 4.1., 4.2., 5.1., 6.1., 8.1., 8.2., 8.3., 15.1, 16.2.	a, b, c, e, f, g, i, j, o, p, r, s, t

Outcome expectations

reduce pain catastrophizing beliefs about engaging in ACB using CBT strategies	PO.9, PO.10, PO.11, PO.13	1-9	Information; self-regulation; SMART goal setting; graded tasks; actioning planning; modelling; persuasion; personalising risks and benefits; barrier / facilitator identification; modelling; active participation;	1.1, 1.2.,1.4., 2.4. 3.1., 4.1., 4.2., 5.1., 6.1., 8.1., 8.2., 8.3., 11.2, 15.1, 16.2.	a, b, c, e, f, g, i, j, o, p, r, s, t
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Emotion

reduce fear of engaging in ACB using CBT strategies	PO.9, PO.11, PO.14,	2, 6	Relaxation training; thought challenging, cognitive re-framing; barrier / facilitator identification; re-cap training	1.2., 1.6., 2.4, 4.1 4.2., 5.1., 6.1., 8.1., 8.2., 8.3., 11.2.,	a, b, c, e, f, g, i, j, o, p, r, s, t
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Skills

develop coping skills using the CBT strategies and tools provided	PO.10, PO.11, PO.13	1-8	Relaxation training; action planning; attention management skills; communication skills; thought challenging; problem-solving skills; communication skills; future planning	1.1.,1.2., 1.4., 1.6., 4.1., 4.2., 5.1., 6.1., 8.1.,8.2., 8.3., 11.2.,	a, b, c, e, f, g, i, j, o, p, r, s, t
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Self-regulation

develop ability to set goals, graded tasks and monitor progress using the tools provided	PO.11, PO.12, PO.13, PO.14,	2-8	Information; self-regulation; SMART goal setting; graded tasks; action planning; modelling; persuasion; barrier / facilitator identification; modelling; implementation intentions	1.1, 1.4., 1.6., 2.4., 3.1., 4.1., 4.2., 5.1., 6.1. 8.1. 8.2, 8.3., 11.2.	a, b, c, e, f, g, i, j, o, p, r, s, t
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Behavioural goals					
use behaviour regulation tools to set goals and monitor progress	PO.10, PO.12, PO.13,	1-8	SMART goal setting; self-monitoring; action planning; barrier / facilitator identification	1.1, 1.4., 1.6., 2.4., 3.1., 4.1., 4.2., 5.1., 6.1. 8.1. 8.2, 8.3.,	a, b, c, e, f, g, i, j, o, p, r, s, t
<hr/>					
Behavioural capability					
demonstrates retention of skills training using tools provided	PO.10, PO.12, PO.13,		Faction information: self-regulation; SMART goal setting; graded tasks; actioning planning; modelling; persuasion; barrier / facilitator identification; modelling; personalising risk	1.1, 1.4., 1.6., 2.4., 3.1., 4.1., 4.2., 5.1., 6.1. 8.1. 8.2, 8.3.,	a, b, c, e, f, g, i, j, o, p, r, s, t
<hr/>					
Social support					
use tools to develop communication skills / problem solve; identify sources of support;	PO.11, PO.12, PO.14,	7-8	Information; barrier / facilitator identification; automatic and personalized feedback / communication skills training; re-cap training	1.1, 1.4., 2.4., 3.1., 4.1., 4.2., 5.1., 6.1. 8.1. 8.2, 8.3., 11.2.,	a, b, c, e, f, g, i, j, o, p, r, s, t
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Facilitation

engage in future planning; use tools to develop problem solving skills; become aware of typical barriers and facilitators of pain SM	PO.9, PO.12;	7-8	Faction information: action planning; modelling; persuasion; barrier / facilitator identification; modelling; active participation; personalising risk; implementation intentions; Problem solving e.g. BEST acronym;	1.1, 1.4., 1.6., 2.4., 3.1., 4.1., 4.2., 5.1., 8.2, 8.3.,	a, b, c, e, f, g, i, j, o, p, r, s, t
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* see Mode of delivery coding scheme: Table 5.10 for detailed description of modes of delivery

Appendix 3 Performance objectives

Table 2.4

Desired change and performance objectives of the Feeling Better intervention

Change objective 1:	Enhance participants self-efficacy by end of programme and 3-month follow-up
Performance objective 1	Children accept the importance of pain SM
Performance objective 2	Children become aware of current pain SM behaviour
Performance objective 3	Children identify barriers and facilitators to be addressed
Performance objective 4	Children identify ACBs (sub-behaviours) to be improved
Performance objective 5	Children understand how and when to use CBT strategies to cope with pain
Performance objective 6	Children understand how to monitor progress in ACBs (sub-behaviours)
Performance objective 7	Children identify realistic goals: SMART goals
Performance objective 8	Children decide to use relevant CBT strategies to manage pain
Change objective 2:	Increase participants' use of CBT strategies by end of programme and 3-month follow-up
Performance objective 9	Children cope with barriers and use resources
Performance objective 10	Children select appropriate CBT strategies to support functioning despite pain
Performance objective 11	Children seek social support
Performance objective 12	Children work toward graded, realistic goals
Performance objective 13	Children use self-regulation strategies to support practice and recall skills training

ACB: adaptive coping behaviour(s); CBT: cognitive behavioural therapy; SM: self-management; SMART: Specific, Measurable, Attainable, Realistic and Timely

Appendix 4 Systematic review protocol as published in BMC Systematic Reviews

The effectiveness of information and communication technology-based psychological interventions for pediatric chronic pain: Protocol for a systematic review, meta-analysis and content coding of behaviour change techniques

Angeline Traynor¹, Eimear Morrissey¹, Jonathan Egan¹ and Brian E. McGuire^{1,2}

¹ School of Psychology, National University of Ireland, Galway, Ireland and ² Centre for Pain Research, National University of Ireland, Galway, Ireland.

Abstract

Background: Resource and geographic barriers are commonly cited constraints preventing uptake of psychological treatment for chronic pain management. For adults there is some evidence to support the use of information and communication technology (ICT) as a mode of treatment delivery. However, mixed findings have been reported for the effectiveness and acceptability of psychological interventions delivered using information and communication technology for children and adolescents. This is a protocol for a review that aims to (i) evaluate the effectiveness of psychological interventions delivered using information and communication technology for children and adolescents with chronic pain and (ii) identify the intervention components and usability factors in technology-based treatments associated with behaviour change.

Methods/Design: We will conduct a systematic review to evaluate the effectiveness of psychological interventions for pediatric chronic pain delivered using information and communication technology (ICT). We plan to directly compare ICT-based, psychological interventions with active control, treatment as usual or waiting list control conditions. This systematic review will be reported in line with the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) guidance. Published and unpublished randomized controlled trials will be included and the literature search will comprise Ovid MEDLINE, Ovid Embase, PsycINFO, the Cochrane Library on Wiley, including CENTRAL and Cochrane Database of Systematic Reviews. Grey literature including theses, dissertations, technical and research reports will also be examined. Two review authors will independently conduct study selection, relevant data extraction and assessment of methodological quality. Risk of bias in included studies will be assessed using the Cochrane Collaboration risk of bias tool criteria. Two qualified coders will independently code behaviour change techniques according to the behaviour change taxonomy (v1) of 93 hierarchically clustered techniques and a novel coding scheme for mode of delivery and usability factors. A quantitative synthesis will be conducted if appropriate.

Discussion: The findings of this review may offer insight for healthcare professionals working in chronic pain services and to researchers involved in

designing and evaluating information and communication technology-based interventions.

Systematic review registration: This systematic review and meta-analysis is registered with PROSPERO (CRD42016017657).

Keywords: Cognitive behavioural therapy, psychological, Chronic pain, Meta-analysis, Systematic review, Randomized controlled trials, internet, online, information technology

Background

Description of the condition

Chronic pain is defined by the International Association for the Study of Pain (IASP) as ‘an unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage’ (Prepared by the International Association for the Study of Pain, 1986). In practice and across the literature, chronic pain is widely accepted to be pain which persists for a period of three months or more. Among children and adolescents, recent reports suggest it continues to be highly prevalent. Current prevalence rates estimate between 11-38% of children and young people are affected by chronic or persistent pain (King et al., 2011; Perquin et al., 2000; Swain et al., 2014). Pain in childhood is thought to be a major public health concern across Western countries, particularly because prevalence has been found to increase with age and is predictive of persistent pain in adulthood (King et al., 2011; Perquin et al., 2000; Swain et al., 2014). The most commonly reported pediatric pain conditions include headache, abdominal pain, musculoskeletal pain and multiple or widespread pain. Across pain types, prevalence is generally higher in girls and those with lower socioeconomic status (King et al., 2011; Perquin et al., 2000; Swain et al., 2014).

Chronic pain has an enormous impact on the lives of children and on family functioning in general. Approximately 5-15% of those affected by pediatric chronic pain report severe and debilitating levels of pain (Huguet & Miró, 2008; Stanford et al., 2008). High pain levels are associated with extensive and often sustained negative effects on child health and overall quality of life for the child and family. Severe chronic pain interferes with daily functioning (Hunfeld, J. A., Perquin, C. W., Bertina, W., Hazebroek-Kampschreur, A. A., van Suijlekom-Smit, L. W., Koes, B. W., Passchier, 2002), sleep (Fales, J., Palermo, T. M., Law, E. F., & Wilson, 2015; Meltzer et al., 2005), emotion regulation (Kashikar-Zuck, Zafar, et al., 2013), social functioning (Forgeron et al., 2010), school performance and attendance (Tine Vervoort, Logan, Goubert, De Clercq, & Hublet, 2014) and family functioning (Hunfeld et al., 2001; Palermo & Chambers, 2005; Palermo & Eccleston, 2009; Sleed et al., 2005). Psychological therapies have been found to be effective in reducing pain and disability in young people with chronic pain (Fisher et al., 2014; Palermo et al., 2010). However, many children and young people do not have access to psychological services to support pain management. Commonly reported barriers include a lack of access to trained healthcare professionals, financial constraints, geographic barriers and scheduling issues. There is some evidence to suggest technology-facilitated delivery of psychological

interventions may help resolve some of the current health care access issues (Griffith et al., 2006) .

The reach of technology-delivered treatment could be extensive. A recent Quarterly National Household Survey (QNHS; 2015) of internet usage in the home, found that 85% of households in Ireland have access to the internet (Central Statistics Office, 2015). Estimates for Europe and North America range from 74% - 88% and worldwide this figure is approximately 46% (Internet World Stats, 2015). The advantages of online delivery of interventions include increased convenience for users, reduction of health service costs and isolation of users, the provision of timely information, increased user and supplier control of the intervention, and research-related benefits (Griffith et al., 2006). However, little is known about the impact of change in mode of delivery from traditional face to face to technology-based treatment. As pointed out by Keogh, Rosser and Eccleston (2010) there is little guidance on how to translate therapy from traditional, human-mediated delivery to technology-based platforms (Keogh, Rosser, & Eccleston, 2010).

Description of the intervention

Psychological treatment for chronic pain typically involves a combination of evidence-based cognitive and behavioural strategies such as relaxation training, cognitive restructuring, acceptance-based skills, information and social support. Some techniques, for example cognitive restructuring, are likely to improve symptoms by influencing how the individual interprets and attributes meaning and emotion to the sensation of pain. Other strategies aim to reduce muscle tension and physiological arousal and thereby promote more adaptive response to pain symptoms.

Although the number of ICT-based interventions for pediatric populations is increasing (Cushing & Steele, 2010; Gustafson et al., 2012; Magee, Ritterband, Thorndike, Cox, & Borowitz, 2009; Tonya M Palermo, Eccleston, Lewandowski, & C, 2010; L. M. Ritterband et al., 2013; J. Stinson, Wilson, Gill, Yamada, & Holt, 2009), as a mode of treatment-delivery for pain management, technology-based methods are still very much in their infancy. Research indicates children and adolescents may be particularly amenable to technology-delivered treatment given their reported comfort with, and time spent using, digital technologies (Bennett, 2008). Evaluations of existing examples are mixed. Many ICT-interventions focus on adult pain populations (Eccleston, C., Fisher, E., Craig, L., Duggan, G. B., Rosser, B. A. and Keogh, 2014), some demonstrate promising findings (Palermo et al., 2009) and others are exploratory or have yet to be extensively evaluated for their effectiveness (Connelly, Rapoff, Thompson, & Connelly, 2006; Cottrell, Drew, Gibson, Holroyd, & O'Donnell, 2007; J. N. Stinson, McGrath, et al., 2010). Qualitative reports and treatment satisfaction data would seem to support this conclusion, suggesting technology-based treatment delivery is acceptable to young people with chronic conditions (Stinson et al., 2008; Tercyak, Abraham, Graham, Wilson, & Walker, 2009), However, high attrition and low adherence rates are reported across the literature and suggest further evaluation of the importance of

contact with therapist and the acceptability of online or technology-based delivery of treatment is necessary (Cavanagh, 2010; Cushing & Steele, 2010; Eccleston, C., Palermo, T. M., Williams, A. C. D. C., Lewandowski Holley, A., Morley, S., Fisher, E., & Law, 2014; Fisher, E., Law, E., Palermo, T. M., & Eccleston, 2015; L. M. Ritterband et al., 2013). Other than the recent review by Fisher et al. (2015), few studies have evaluated ICT-delivered therapies for the management of pediatric chronic pain compared to traditional face to face therapies. In addition to larger sample sizes and the inclusion of active comparators, Fisher and colleagues call for further investigation that allows a better understanding of effective features of ICT-based interventions (Fisher, E., Law, E., Palermo, T. M., & Eccleston, 2015).

How the intervention might work

Treatment effects may result from the provision of evidence-based psychological treatment and may also be facilitated by the characteristics of treatment delivery. The effectiveness of ICT-delivered treatment may be related to presence and combination of specific behaviour change techniques and how they are implemented or the usability of the platform in terms of efficiency, learnability, satisfaction or personalisation. Health benefits in the form of behaviour change or symptom improvement associated with ICT-interventions may be attributable to an amalgamation of user characteristics, environmental factors and the presence of human support in the form of an e-coach or online therapist (Cheek et al., 2015; Ritterband, 2009). It may also be that the use of engaging and interactive technology in treatment encourages children to perceive therapeutic instruction in terms of discovery. The personification of an e-coach or online therapist in the form of an avatar or friendly image may encourage the perception of the therapist as a facilitator rather than a teacher or figure of authority. Health benefits may also be boosted by the lack of time constraints or unlimited access typically associated with online interventions. The freedom to access the intervention at a convenient time may facilitate the delivery of timely information and promote the therapeutic learning or practice as an ongoing or long-term habit rather than a scheduled classroom or clinic based activity (Bennett, 2008; Chang et al., 2004).

Why it is important to do this review

Chronic pain may present as a result of injury, infection or surgical procedure and often, no apparent cause is found. Despite the personal and economic burden of pediatric chronic pain for the individual and family as a whole, it is often seen as symptom rather than a condition in its own right (Groenewald et al., 2014). Perhaps as a consequence, chronic pain and particularly pediatric chronic pain does not receive the same focus or priority given to other chronic conditions. Efforts are needed to determine how interventions should be developed to address pediatric chronic pain management. It will be important to determine whether appropriate, effective ICT-based pain management interventions can be identified, in order that effective therapies can be developed and distributed to improve pain symptoms in school age children. In the context of a rapidly expanding suite of apps and other ICT-based clinical tools, we believe it is important to determine the acceptability and effectiveness of ICT-delivered therapies compared with

traditional face to face therapies. Identifying the intervention technology components and behaviour change techniques used in current interventions will contribute to the evidence base for the development of with ICT-based, pediatric pain management interventions.

Purpose of the proposed review

The purpose of the proposed systematic review is to evaluate the effectiveness of psychological interventions for pediatric chronic pain delivered using ICT. We plan to directly compare ICT-delivered interventions with active control, treatment as usual or waiting list control conditions. We will assess treatment efficacy based on the PedIMMPACT recommendations (P. J. McGrath et al., 2008) and the recommended IMMPACT criteria outlined for interpreting the clinical importance of treatment outcomes in chronic pain clinical trials (Dworkin et al., 2008). Our primary outcome of interest will be pain interference (i.e., reduced disability), and pain intensity. Our secondary outcomes will be emotional functioning, global rating of improvement, quality of life, adverse events and treatment satisfaction. We hypothesize that ICT-delivered interventions will differ in terms of treatment benefits for both our primary and secondary outcome measures of interests. This review protocol parallels that of Eccleston, Fisher, Craig, Duggan, Rosser and Keogh (Eccleston, C., Fisher, E., Craig, L., Duggan, G. B., Rosser, B. A. and Keogh, 2014; Eccleston C, Keogh E, Duggan GB, 2012) which focuses on telemedicine for chronic pain management in adults. It also builds on the recently published systematic review by Fisher et al, (2015) which focuses on remote delivery of psychological therapies for pediatric chronic pain in children and adolescents. The proposed review will go beyond previous evidence syntheses by exploring putative intervention components and usability factors which may act as potential sources of variation in effects.

Aim

This systematic review will evaluate the features and effectiveness of psychological interventions for pediatric chronic pain delivered using ICT.

Key Objectives

The key objectives of this study are the following:

1. To evaluate the effectiveness of psychological interventions for pediatric chronic pain delivered using ICT in comparison with active control, treatment as usual or waiting list control conditions.
2. To identify the intervention components (theoretical basis, behaviour change techniques, interactive elements and level of human support) associated with effectiveness in ICT-based interventions relative to active control, treatment as usual or waiting list control conditions.

Methods

Criteria for considering studies for this review

The conduct and report of the proposed review and meta-analysis will adhere to the reporting guidelines of the “Preferred Reporting Items for Systematic Reviews and Meta-analyses Protocols (PRISMA-P) 2015 Statement” and the PRISMA statement (see Additional file 1) (Moher D, Liberati A, Tetzlaff J, Altman DG, 2009; Moher D, Shamseer L, Clarke M, Ghersi D, Liberati A, Petticrew M, Shekelle P, 2015). This systematic review protocol was registered with the International Prospective Register of Systematic Reviews (PROSPERO) database (CRD42016017657).

Types of studies

We will assess all published and unpublished randomised controlled trials (RCTs) which evaluate the effectiveness of ICT-based psychological interventions for pediatric chronic pain management. This will include treatment groups compared with active control, treatment as usual, or waiting list control.

Type of participants

We will focus on children and adolescents 18 years of age and younger that meet the criteria for a diagnosis of non-malignant chronic pain. In the absence of a clinical diagnosis, chronic pain will be defined as self-reported persistent pain lasting 3 months or more. This is consistent with the definition of chronic pain provided by the International Association for the Study of Pain, which states: pain is an ‘unpleasant sensory and emotional experience, associated with actual or potential tissue damage, or described in terms of such damage’ (Prepared by the International Association for the Study of Pain, 1986). Chronic pain conditions may include headache or migraine, pain in any body area and pain associated with a range of conditions (e.g. rheumatoid arthritis, myofascial pain conditions, neuralgia, fibromyalgia). Studies that enrolled adults or patients who had been experiencing pain for less than the 3-month threshold duration will be excluded from the present review.

Type of interventions

We will include interventions which evaluate the effect of psychological treatment for chronic pain in children and adolescents, delivered using ICT. At least one arm of each included trial must involve a predominantly psychological therapy or include definable psychotherapeutic content. In line with McGuire et al., (2014), psychological interventions will be included if they use techniques often used for chronic pain management including relaxation training, cognitive restructuring (i.e. changing pain-related beliefs, reducing catastrophic thinking, etc.), setting and working towards behavioural goals (e.g., exercise), behavioural activation, and problem-solving. Studies that use methods such as meditation, mindfulness, stress management or other techniques to improve pain self-management will be included. All interventions must aim to reduce pain characteristics, functional limitations, psychological distress and/or more adaptive behaviour change. A measure of pain characteristics must be included in any examination of the effects

of psychological treatment on multiple outcomes (see *Types of outcomes* section below).

Types of intervention delivery

Studies must evaluate ICT-based interventions which function as the primary mode of treatment delivery. Interventions which involve the support of a health care professional will only be considered if the primary mode of treatment delivery is ICT-based. Studies where ICT is used to facilitate traditional treatment but does function as the primary source of treatment (e.g. aid symptom monitoring or communication only) will be excluded. No restriction will be placed on the level of user interaction or data input in a given ICT-based, intervention platform. We will include studies that evaluate any information and communication-based intervention delivered in the home, school or community.

Type of outcome measures

Primary outcomes

The primary outcomes of interest are those that measure change in pain characteristics including intensity, severity or frequency and pain interference including physical and social interference (e.g. related school absenteeism) from pre to post treatment. Pain intensity, severity and frequency are generally measured using self-reported verbal or numerical rating scales or visual analogue scales. Pain interference is commonly measured using psychometric tools with established validity and reliability e.g. Functional Disability Inventory (FDI) (Kashikar-Zuck et al., 2011) or the Pediatric Quality of Life Inventory (PedsQL) (Varni, Seid, & Kurtin, 2001). In cases where the chronic pain condition refers to chronic headache pain or migraine, psychometric tools such as the PedMIDAS (Hershey, A. D., Powers, S. W., Vockell, A. L., LeCates, S., Kabbouche, M. A., & Maynard, 2001) may be used. As per Eccleston et al., (2012) and McGuire et al., (2014), we will also report the responder rate (the percentage of subjects in the treatment group with at least 50% reduction in the primary efficacy measure)(Eccleston Fisher E, Craig L, Duggan GB, Rosser BA,Keogh E., 2012; McGuire et al., 2014).

Secondary outcomes

Secondary outcomes of interest include emotional functioning (self-reported measures of psychological distress such as depression and/or anxiety), quality of life (self-report questionnaires assessing the impact of chronic pain on quality of life) and global ratings of improvement (self-reported measure of change in subjective sense of wellbeing). Data relating to treatment acceptability and satisfaction, retention and attrition will also be extracted as a secondary outcome. Adverse events will also be reported. These variable may be measured using psychometric tools with established validity and reliability such as the Pediatric Quality of Life Inventory (PedsQL) (Varni et al., 2001), the Children's Depression Inventory (CDI) (Kovacs M., 1981). Finally, we will include variables which are measured as a discrete outcome or as a sub-scale within a composite measure.

Search methods for identification of studies

No restrictions will be placed on the date of publication or publication status. Studies will be included if the full report is accessible in English, either through electronic search or through contact with the author.

We will design and conduct a three step search strategy using methods recommended by the Cochrane Collaboration (Higgins JPT, 2011).

1. The initial search strategy will be designed with consideration of other similar reviews' strategies. In addition, we will conduct an initial search of journals indexed in MEDLINE and PsycINFO with the aim of extracting and compiling a comprehensive list of text or key words contained in the title, abstract and subject descriptors/MeSH terms of relevant articles. All identified key words and their synonyms will be compiled and used to develop an individual search strategy for MEDLINE. This will be revised appropriately for each database searched (see Table 1).

2. We will search several databases including: OVID MEDLINE®, OVID EMBASE, OVID PsycINFO and the Cochrane Central Register of Controlled Trials (CENTRAL). The databases will be searched for randomised controlled trials of ICT-delivered interventions for pediatric chronic pain conditions through the time period of database inception to the present.

Clinical trial registries will be searched to identify completed and in-progress trials. This will include the following databases:

- ClinicalTrials.gov (clinicaltrials.gov),
- The metaRegister of controlled trials (mRCT), (controlled-trials.com)
- The World Health Organization (WHO) International Clinical Trials Registry Platform (ICTRP) (www.who.int/ictrp/en/) for trials.

Grey literature including theses, dissertations, research and technical reports and conference papers will also be examined. Grey literature source selection will be guided by CADTH's Grey matters: a practical search tool for evidence-based medicine (Canadian Agency for Drugs and Technologies in Health, 2011). This will include the following databases:

- Scopus (www.scopus.com);
- ProQuest (www.proquest.com);
- Ethos (<http://ethos.bl.uk>);
- Open Grey (www.opengrey.eu/ OpenGrey);

- TRIP (Turning Research into Practice <http://www.tripdatabase.com>);
- WorldCat (www.worldcat.org);
- National Technical Information Service (NTIS, <http://www.ntis.gov/>).

3. The initial electronic search strategy will be supplemented by screening the reference lists of included reports and articles to identify additional studies. If not established through other methods, authors will be contacted for details regarding the status of a given study.

Table 1: Addition file 2: Details of Search Strategy

Data collection and analysis

Selection of studies

The titles and abstracts of publications obtained by the search strategy will be independently screened by two authors (AT, BMG). Those that fail to meet the outlined inclusion criteria will be removed. All remaining publications will be retrieved for further scrutiny. Two review authors (AT, BMG) will independently assess the full text of studies which initially meet the review criteria. Disagreements between review authors will be discussed until resolved; in the event a resolution cannot be reached a third review author will arbitrate (JE). A record will be kept of all articles excluded at this stage and the reason for their exclusion. We will produce a PRISMA flow diagram to illustrate the search and systematic review process as recommended in Chapter 6 of the Cochrane Handbook (Higgins JPT, 2011).

Data extraction and management

A data extraction form will be created prior to data extraction. Data will be extracted independently by one reviewer (AT) and verified by another (BMG) using a customised form, which will be piloted prior to use. Disagreements in data extraction will be resolved through discussion with the primary data extractor, as required. In the event that resolution cannot be reached, a third review author will arbitrate (JE). The finalised data will be entered into RevMan 5.3 (The Nordic Cochrane Centre, 2014). Multiple publications of the same study will be identified, linked and used for all relevant reported data. In such cases, the original publication will be given priority. Where the necessary outcome data are unavailable, we will contact study authors. If the data remains unavailable, the study will not be included in any assessment. The authors will not be blind to the study author, institution or journal.

We will extract data relevant to the following categories: (i) Study population and design, (ii) Intervention and (iii) Outcome. Characteristics of included studies' table(s) will be created and may include the following information:

- Participant characteristics
- Geographic location
- Assessment periods
- Description of providers of intervention and comparison interventions
- Primary and secondary outcomes
- Theoretical basis (domains identified)
- Therapeutic content (characteristics of psychological therapies)
- Mode of delivery (Internet, smartphone app, telephone, text)
- Behaviour change techniques
- Control condition
- Intensity (e.g. no. of sessions, total contact time, duration)
- Treatment engagement (retention and attrition)

The use of theory in the included interventions will be coded according to the Theoretical Domains Framework (TDF) (Cane, O'Connor, & Michie, 2012). This is an integrative framework which was developed and validated by Cane, O'Connor and Michie (2012). The TDF summarises the range of psychological theory potentially driving behaviour change, into a total of 14 distinct domains. In line with the approach taken by Little et al (2015), descriptions of intervention and control conditions will be assessed to determine if and to what extent TDF domains are targeted within (Little Elizabeth, Preece & Eccles, 2015). This process will be carried out independently by two coders (AT, EM) using a data extraction form similar to the form used by Little et al (2015). Inter-rater reliability will be calculated and discrepancies will be discussed until resolved.

The behaviour change technique taxonomy (v1) of 93 hierarchical clustered techniques (Michie et al., 2013) will be used to code intervention content. Mode of delivery and usability factors will be coded using a novel coding scheme adapted by Webb et al. (2010) and van Genugten (2016) (& Michie, 2010; L. van Genugten, E. Dusseldorp, T.L. Webb, 2016). Two qualified coders (AT, EM) will independently code the behaviour change techniques described in the intervention and control conditions. Kappa and percentage disagreement will be calculated. Disagreements between reviewers will be discussed until resolved or with third party arbitration (BMG) if required.

Assessment of risk of bias

For each included study the review authors (AT, BMG) will independently carry out a domain-specific assessment of risk of bias using the recommended Cochrane Collaboration's tool for risk of bias assessment (Higgins JPT, 2011). This will

involve the classification of risk of bias in included studies as ‘low’, ‘unclear’ or ‘high’ risk of bias. If the authors (AT, BMG) disagree the final rating will be made by consensus with the third author (JE). The domains assessed will include:

- Random sequence generation - to assess the potential for selection bias
- Random allocation concealment - to assess the potential for selection bias
- Blinding of participants and personnel - to assess the potential for performance bias (both participants and outcome assessors)
- Blinding of outcome assessment - to assess the potential for detection bias
- Incomplete outcome data - to assess the potential for attrition bias
- Selective reporting - to assess the potential for reporting bias
- Other bias - to assess the potential for other sources of bias not covered in other domains

Where necessary, we will contact the study authors to request missing data and/or data clarification. The quality of the data included in the review and the presence of any serious flaws will be reported.

Overall quality of the evidence

If appropriate, we will use the Grading of Recommendations Assessment, Development and Evaluation (GRADE) approach to summarise the quality of evidence for each outcome at post-treatment and follow-up (Balshem, H., Helfand, M., Schunemann, H. J., Oxman, A. D., Kunz, R., Brozek, J., . . . Guyatt, 2011; Higgins JPT, 2011; Mustafa et al., 2013). We will use GRADEprofiler (GRADEpro) to import data from Rev Man 5.3 to create Summary of Findings table(s) (Guyatt, Oxman, et al., 2016; Guyatt, Thorlund, et al., 2016; Higgins JPT, 2011). This will report outcome-specific information concerning the overall quality of evidence (risk of bias, inconsistency, imprecision and indirectness). Only the most important outcomes will be included in each ‘Summary of Findings’ table. As per Fisher et al., (2015), the most important outcomes will be deemed those with the largest number of participants in each arm (Fisher, E., Law, E., Palermo, T. M., & Eccleston, 2015).

Sensitivity analysis

Sensitivity analysis based on methodological rigour and risk of bias will be carried out to determine the robustness of results. Studies deemed to be of high or unclear risk of bias across different domains will be systematically excluded then included, in order to assess differences in the overall effect estimates. If no significant differences exist, the studies will be included in the main analysis.

Measures of treatment effect

In order to synthesise data across studies, we will compute and report mean differences where identical scales are used to measure the same clinical outcome. Where different scales are used to measure the same clinical outcome, we will compute standardised mean differences (SMDs) (otherwise weighted mean differences). For dichotomous data, we will report odds ratios (ORs), 95% confidence intervals (CIs) and number needed to treat to benefit (NNTB). For continuous data we will most likely report standardised mean differences and 95% confidence intervals. There are a small number of studies in this area. We expect data to be sparse, event rates may be low or study size may be small therefore we will use Mantel-Haenszel methods in analyses of dichotomous data. We anticipate effect sizes will be similar but not identical across studies therefore a random-effects model will be used in analyses. Pain related interference and pain intensity outcomes tend to be reported using primarily continuous data and studies which include headache conditions are likely to report pain symptoms using primarily dichotomous data. Chronic pain conditions will be categorised according to pain condition. In line with Fisher et al (2015), data from studies reporting mixed pain conditions will be entered into all appropriate analyses. When studies use more than one measure for a given outcome, we will extract the most reliable or widely accepted. As per Eccleston et al (2014) and Fisher et al (2015), the timeframe allowed for collection of follow-up data will range from three to twelve months' post treatment. If more than one-time point is available at follow-up, the latest data collection point will be extracted.

Missing Data

Where necessary, attempts will be made to contact the lead authors of included studies to request missing data. We will, where necessary, calculate standard deviations using the methods described in the Cochrane Handbook for Systematic Reviews of Interventions (2011). Included studies will be scanned for other statistics including confidence intervals, standard errors or *p* values that would allow for its calculation. If missing data required for analyses cannot be obtained from the study author or extrapolated from other statistics, the study will be excluded. We will record the use of intention to treat analyses (ITT) and if sufficient data is available we will conduct sub-group analyses to evaluate the impact of inclusion or exclusion of non-completers in final study analysis.

We will report rates of missing outcome data per arm and refer to the Cochrane risk of bias tool for missing outcome data in any evaluation of imputation methods. Finally, we address the potential impact of missing data on the findings of the review in the Discussion section (Higgins JPT, 2011).

Unit of analysis issues

We anticipate unit of analysis issues such as repeated observations of the same outcome and studies including multiple intervention arms. For studies reporting repeated measurements of the same outcome, we will extract data at the following time points: baseline, post-treatment (not longer than 3 months post-randomisation), and follow-up (not longer than 12 months post-randomisation). For studies including more than two intervention groups, we will adhere the recommended method suggested by the Cochrane Collaboration in section 16.5 for

combining multiple groups from one study (Higgins JPT, 2011). If cluster-randomised controlled trials are included, we will check for unit of analysis errors. Where possible, we will recalculate results using the appropriate unit of analysis (Higgins 2011). As per the Eccleston et al., (2012) protocol, all psychological intervention conditions will be labelled ‘treatment’ and all comparator conditions will be labelled ‘control’ conditions.

Assessing for heterogeneity

We will assess heterogeneity by calculating Chi^2 and I^2 values for all outcome variables. Statistical heterogeneity will be considered substantial if I^2 values are above 50%. We will also assess the impact of heterogeneity through sensitivity analyses and assume the appropriate random-effects or fixed-effect model in meta-analyses accordingly (Higgins JPT, 2011).

Assessment of reporting biases

According to section 10.1 of the Cochrane Handbook, reporting biases arise when dissemination of findings is influenced by the nature and direction of results (Higgins JPT, 2011). For this reason, we will, where possible, retrieve and compare the protocol for the included studies with the final reports.

The potential for small study effects such as publication bias will be assessed visually by inspection of funnel plots of estimated effects by standard error and using statistical tests which are in line with recent recommendations (Egger, M., Smith, G. D., Schneider, M., & Minder, 1997; Sterne et al., 2011). Funnel plots will be assessed if ten or more studies are identified. The possible reasons for asymmetry will be investigated.

Data synthesis

We will pool data using the Cochrane Collaboration’s Review Manager Software, RevMan 5.3 (RevMan 2014). A quantitative synthesis will be carried out only if the included studies are sufficiently homogenous in terms of quality, study design, participants, interventions, outcomes and type of analyses to provide a meaningful summary of effects. A narrative synthesis will be carried out if there is insufficient data to justify a formal meta-analysis. For continuous data we will calculate and report standardised mean differences (SMDs) and 95% confidence intervals (CIs). For dichotomous data we will calculate and report odds ratios (ORs), 95% confidence intervals (CIs) and number needed to treat to benefit (NNTB). In the event that data is sparse, we will use Mantel-Haenszel methods in analyses of dichotomous outcomes. Given the likely differences in interventions, comparators and participants we expect sufficient clinical heterogeneity that included studies will estimate different but related intervention effects. As some heterogeneity is inevitable we anticipate a random effects model will be used in analyses.

In line with Little et al (2015), a Pearson correlation (two-tailed) will be used to explore the relationship between the total number and frequency of different TDF

domains coded and the effect size of the intervention for both the ICT-based interventions and the control conditions. For example, the number of different domains coded in the control group will be subtracted from the number of different domains coded in the intervention group. Sensitivity analysis will be used to investigate whether subtracting domains that appear in the control group impacts on the findings (Little Elizabeth, Presseau & Eccles, 2015). If there are no significant differences, data synthesis will be descriptive e.g. the proportion of studies that target specific domains will be identified.

Subgroup analysis and investigation of heterogeneity

If sufficient data are available, several subgroup analyses will be performed. The following factors will be examined in sub-group analyses to determine their effect on the response to a psychological intervention for pediatric chronic pain:

- Technology type - differences between modes of delivery including text messaging, online websites, virtual or game-based programmes, smartphone applications and telephone-based treatment delivery methods.
- Contact with therapist - the degree of guidance which features in each intervention, ranging from 'pure' and unsupported to 'guided' and frequently supported self-management interventions.
- Pain type - differences between pain-type including arthritis, back-pain, abdominal pain, mixed pain and headache pain.
- Behaviour change techniques used (based on the findings of previous studies which suggest specific techniques are associated with effectiveness)
- Usability factors used (based on the findings of previous studies which suggest specific factors are associated with effectiveness)

Of these subgroup analyses, modes of delivery and the extent of personalised contact with the therapist may be the most important because it remains unknown which ICT-based intervention types are most effective for pediatric chronic pain management and also how effective personalised contact with therapist (e.g. an e-coach) is in comparison with pure (e.g. no contact with therapist) self-led programmes.

Discussion

ICT-based psychological interventions may have the potential to address both the pain and disability associated with chronic pain conditions and the resource and geographic barriers to uptake of psychological treatment for chronic pain management. Reviews which identify effective components of ICT-based interventions have tended to focus on physical activity (Bird EL, Baker G, Mutrie N, Ogilvie D, Sahlqvist S, 2013) or multiple behaviours (& Michie, 2010), but to date these have not focused on chronic pain populations. This review will be the first to our knowledge, to evaluate the components, usability and effectiveness of ICT-based psychological interventions for children and adolescents with chronic

pain. The findings of this review will offer insight for those involved in the design and development of complex psychological and technology-based interventions.

Limitations

The findings from the current study will have certain limitations. First we anticipate a small number of studies will be included. Second, it is expected that some interventions will fail to provide a detailed description of intervention content or to report explicit use of theory. This is a limitation of retrospective content coding. To address this issue, the lead authors of the included papers will be contacted and asked to provide more information. Also, content analyses will be conducted independently by two reviewers in an effort to enhance the reliability of the extracted data.

Abbreviations

MEDLINE: Medical Literature Analysis and Retrieval System Online Abbreviations; EMBASE: Excerpta Medica database; PRISMA: Preferred Reporting Items for Systematic Reviews and Meta-Analyses; RCT: randomised controlled trials; GRADE: Grades of Recommendation Assessment, Development and Evaluation; IASP: International Association for the Study of Pain; JIA: juvenile idiopathic arthritis; OR: odds ratio; MeSH: Medical Subject Headings; mRCT: metaRegister of Controlled Trials; PRISMA: Preferred Reporting Items for Systematic Reviews and Meta-Analyses; PRISMA-P: Preferred Reporting Items for Systematic Reviews and Meta-Analyses protocols 2015 statement; WHO ICTRP: World Health Organization International Clinical Trials Registry Platform; BMG: Brian E. McGuire; AT: Angeline Traynor; JE: Jonathan Egan; EM: Eimear Morrissey.

Additional file

Addition file 1: Populated PRISMA-P checklist (DOC 75 kb)

Ethical Approval and Consent to participate

Not applicable

Consent for publication

Not applicable

Competing interests

The authors declare that they have no competing interests.

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Availability of data and materials

Not applicable

Authors' contributions

AT: participated in the conception and design of the study, developed the search strategy, carried out the initial background search and drafted the manuscript. BMG: was involved in the conception and design of the study, refinement of the search strategy, inputting on methodology and intellectual property and final critical review of the manuscript. JE: contributed to the conception and design of the project and refinement of the search strategy. EM: provided input on the methodology. All authors read and approved the final manuscript.

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Not applicable

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Appendix 5: Systematic review search strategy

Design

- 1 randomized controlled trial.pt.
- 2 controlled clinical trial.pt.
- 3 randomized.ab.
- 4 placebo.ab.
- 5 trial.ti.
- 6 clinical trials as topic.sh.
- 7 randomly.ab.
- 8 trial.ti.
- 9 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8

1 exp animals/ not humans.sh.

1 9 not 10

Populations

1 exp Child/

1 exp infant/

1 exp Adolescent/

1 (child or adolescent or infant or juvenile or pediatric or paediatric
5 or young person or young people or youth or young adult or teen or
teenager or boy* or girl* or schoolchild).mp.

1 12 or 13 or 14 or 15

Interventions

1 exp Psychology/

1 exp Psychotherapy/

1 exp Behavior Therapy/

2 (((psycholog* or behavior) and therap*) or hypnos* or relaxation
0 or family or mindfulness or meditation or acceptance or imagery or
cogniti* or psychotherp*).mp.

2 17 or 18 or 19 or 20

2 exp Internet/

2 exp Telecommunications/

2 (telemedicine or tele-medicine).mp.

2 (telehealth or tele-health).mp.

2 (e-health or ehealth).mp.
 6
 2 (((internet* or world) and wide and web) or www or web-based or
 7 email or e-mail* or online or social media).mp.
 2 ((health* or therap* or intervention* or treat* or assist* or
 8 selfmanag* or self- manag*) adj6 (computer* or technolog* or
 software)).mp.
 2 (telephone or smartphone or cellphone* or mobile or mobile health
 9 or mhealth or m-health or text* or app).mp.
 3 ((ICT or information) and communication technology).mp.
 0
 3 ((inform* or communicat* or interact*) adj6 (computer* or
 1 technolog* or software)).mp.
 3 (virtual reality or augmented reality or VR or AR or game based or
 2 gamification).mp.
 3 30 or 31 or 32 or 33 or 34 or 35 or 36 or 37 or 38 or 39 or 40
 3

Outcomes

3 exp Pain/
 4
 3 (chronic pain or recurrent pain or persistent pain or nociceptive
 5 pain or psychogenic pain or neuropathic pain or somatic pain).mp.
 3 (pain or headache or migraine or cephalagi or neuralgi).mp.
 6
 3 (arthritis or juvenile idiopathic arthritis or JIA or juvenile
 7 fibromyalgia syndrome or fibromyalgia or ankylosing spondylitis
 or juvenile spondylitis or rheumatoid arthritis or osteomyelitis).mp.
 3 (chronic pelvic pain or non-cardiac chest pain or complex regional
 8 pain syndrome or mixed pain or neuropathic pain or mixed pain or
 musculoskeletal pain or knee pain or back pain or low back
 pain).mp.
 3 (stomach ache or tummy ache or abdominal pain or belly ache or
 9 recurrent abdominal pain).mp.
 4 (((ear ache or odontalgi or myofascial) and pain) or orofacial pain
 0 or facial) and pain).mp.
 4 22 or 23 or 24 or 25 or 26 or 27 or 28
 1
 4 11 and 16 and 21 and 29 and 41
 2

Appendix 6 Data Extraction Form

1. Study ID [textbox]
 2. Authors [textbox]
 3. Year of publication / Citation [textbox]
 4. Publication status (published or unpublished) [textbox]
 5. Title of article [textbox]
 6. Study design [textbox]
 7. Design [textbox]
-

- a. Recruitment period [textbox]

Complete description of treatment condition

- b. Nature of intervention: [textbox]
- c. Exposure - number of sessions: [textbox]
- d. Assessment - duration of follow up: [textbox]
- e. Attrition –pre-enrolment, post-treatment and at follow-up, across trial arms [textbox]

Complete description of control condition

- f. Nature of intervention: [textbox]
- g. Exposure - number of sessions: [textbox]
- h. Assessment - duration of follow up: [textbox]
- i. Attrition –pre-enrolment, post-treatment and at follow-up, across trial arms [textbox]

8. Study Characteristics

- a. Eligibility
 - i. Inclusion / exclusion criteria [textbox]
- b. Participants
 - i. Age (mean and SD) [textbox]
 - ii. Sex [textbox]
 - iii. Source / Recruitment [textbox]
 - iv. Diagnosis [textbox]

- v. Mean years of pain [textbox]
- vi. Number randomised to intervention group
 - a. End of treatment [textbox]
 - b. Start of treatment [textbox]
- vii. Number randomised to control group
 - a. End of treatment [textbox]
 - b. Start of treatment [textbox]

9. Measures reported?

- a. Primary measures [textbox]
- b. Secondary measures [textbox]
- c. Measures of interest to this review [textbox]

10. Outcomes:

- 1. Results for outcomes: [table]
 - a) Results for outcomes at each assessment point –
 - i. Effects
 - ii. Variance
 - iii. Statistical significance for differences between groups.
 - b) Was there a statistically significant effect on outcomes?

* $p > 0.05$, ** $p > 0.01$, *** $p > 0.001$

11. Theoretical Basis [multiple choice / text box]

12. Intervention description

- a. Components - elements of intervention (if mentioned): [textbox]

13. Description of intervention provider / therapist [textbox]

14. Intervention modes delivery [multiple choice]

Appendix 7: Coding with the Theoretical Domains Framework

Study	Description of intervention	Domain	Construct	Determinant	Description of control	Domain	Construct	Determinant
Connelly (2006)	<p>Participants in the treatment group continued to follow the recommendations of their neurologist and were sent the Headstrong CD-ROMs immediately following the baseline period.</p> <p>Module number 1: Education</p> <ol style="list-style-type: none"> 1. How to use the Headstrong Program 2. Types of headache 3. Prevalence of headache 4. Typical features of headache 5. How headache is diagnosed and treated 6. The headache pain puzzle <p>Homework: Submit record sheets (password and quiz sheet) Complete headache triggers assignment</p> <p>Module number 2: Relaxation</p> <ol style="list-style-type: none"> 1. Rationale for relaxation 2. How to use guided imagery 3. How to use deep breathing 4. How to use progressive muscle relaxation <p>Homework: Submit record sheets Submit logs of relaxation</p>	10. Memory Attention and Decision Processes	Attention	Attention focused through intervention	<p>Participants in the wait-list control group continued to submit records weekly while following the recommendations of their neurologist.</p> <p>Such recommendations consisted of medication prescriptions (see Table 2 for breakdown of medications prescribed) and dietary and sleep schedule advice.</p> <p>Participants in the wait-list control condition were contacted weekly to encourage consistent record-keeping and to ensure there was no systematic bias in the level of therapist attention received by both groups.</p> <p>The wait-list period continued for 2 months, at which point children were offered the opportunity to receive the Headstrong program and followed the procedures outlined for the treatment group.</p>	10. Memory, attention, decision processes	Attention	Weekly calls (Attention focused through record keeping)
		11.Env context & resources	Material resources	CD-ROM		1.Knowledge	Procedural knowledge	Neurologist recommendations / Advice: (dietary advice and sleep schedule)
		1.Knowledge	Knowledge	CD-ROM – Education module 1 (Education: types, prevalence, diagnosis info)		12. Social Influence	Social support	Weekly calls
		1. Knowledge	Knowledge	CD-ROM – Education module 2 (Rationale for relaxation)				
		1 Knowledge	Procedural knowledge	CD-ROM – Relaxation				

	<p>Practice</p> <p>Module number 3: 1. Rationale for coping 2. Thought-changing 3. Problem-solving Homework: Submit record sheets Submit thought-changing and problem-solving worksheets</p> <p>Module number 4: Behavior 1. Positive and negative pain behaviors 2. Pain behaviour management 3. Review of all lessons Homework: Submit record sheets Submit pain behaviour management plan worksheet</p> <p>Module 1: The educational module of the Headstrong program consisted of several</p>	<p>2. Skills</p> <p>1. Knowledge</p> <p>4. Beliefs about capabilities</p> <p>6. Beliefs about consequences</p> <p>2. Skills</p> <p>14. Behaviour regulation</p> <p>2. Skills</p> <p>10. Memory Attention and Decision Processes</p>	<p>Skills development</p> <p>Knowledge</p> <p>Perceived behavioural control Outcome expectancies</p> <p>Skills development</p> <p>Action planning</p> <p>Skills development</p> <p>Decision-making</p>	<p>module 2 (“How to.instructions”)</p> <p>CD-ROM – Relaxation module 2 (logs of relaxation practice)</p> <p>CD-ROM – Education module 3 (Rationale for coping)</p> <p>CD-ROM – education module 3 (Thought changing / problem-solving)</p> <p>Worksheets</p> <p>CD-ROM – education module 4 (Pain behaviour modification)</p> <p>Management plan</p> <p>Three different images</p>				
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	<p>fully narrated lessons for which the child would click through in a predetermined order over the course of 1 week.</p> <p>Module 2: The relaxation module consisted of a rationale and subsequent graphic demonstrations and experiential learning of various relaxation techniques (deep breathing, imagery, and progressive muscle relaxation). For imagery, the child could choose among three different images for experiential learning based on his/her preference.</p> <p>Module 3: The thought-changing module consisted of a rationale and interactive means of demonstrating how to change thoughts about common stressful experiences (e.g., academic and social stress as well as headaches) to more helpful thoughts.</p> <p>Module 4: Finally, pain behavior modification primarily required the child to involve the caregiver(s) in devising and implementing an active pain-coping plan based on the skills learned. Graphics, language, and music were selected to be developmentally appropriate, and all components were fully narrated.</p> <p>Further, children were required to complete quizzes and password sheets at the end of each module and submit these via prepaid mail as a means of assessing adherence and use of the content covered in the program.</p>	Behaviour regulation	Self-monitoring	CD-ROM 1-4 Engage with CD-ROM (daily diaries: record sheets, practice logs, adherence, choice of relaxation exercises)					
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	<p>After completion of the Headstrong program, participants submitted daily diaries weekly via prepaid mail for 3 months after treatment. The PedsMIDAS questionnaires were completed and returned at the end of each month.</p> <p>Weekly phone calls continued to ensure consistent record keeping and to corroborate frequency data reported in the diaries.</p>	12. Social Influences	Social support	Weekly calls				
Cottrell (2007)	<p>Telephone calls</p> <p>Manual and tapes</p> <p>Treatment was guided by a counselor in 8 weekly telephone calls that reviewed the learning tasks and addressed any problems encountered by the adolescent. TAT subjects received the STOP Migraines treatment manual that included sections addressing each of the 8 weekly learning tasks. Two relaxation tapes also were provided. The TAT manual was adapted from McGrath and colleagues' manual for pain management and was modified to more specifically address migraine headache management skills (eg, thermal biofeedback training).</p> <p>Week 1</p>	<p>10. Memory attention and decision processes</p> <p>11. Env context & resources –</p> <p>1 Knowledge</p>	<p>Attention</p> <p>Resources</p> <p>Knowledge</p>	<p>Attention focused through phonecalls</p> <p>Tapes</p>	<p>Triptan Treatment—For adolescents with episodic migraine who have not been responsive to NSAIDs and analgesics the medication preference of parents in our setting is typically for an alternate oral acute therapy option, with preventive drug therapy instituted only if triptan therapy fails to adequately control migraines and restore functioning, or is not tolerated or cost effective.</p>	10. Memory Attention and Decision Processes	Attention and Decision making	Oral acute therapy options (decision to engage in research project)

<p>Introduction to program goals, education regarding migraine and tension headache characteristics, discussion of headache triggers, and how to identify and avoid them. Parent/caregiver phone contact: how to best support the participant's effective use of the TAT program.*</p> <p>Week 2 Introduction to recognizing and monitoring headache-related stress; correct and effective acute medication use. Parent/caregiver phone contact: recognizing effective and ineffective coping; rewarding effective rather than ineffective coping.</p> <p>Week 3 Introduction to progressive muscle relaxation and relaxation imagery†; instructed to practice relaxation 20 minutes per day.</p> <p>Week 4 Introduction to relaxation by recall, deep breathing, additional relaxation imagery.† Introduce the application of relaxation skills to preventing, aborting and coping with headaches.</p> <p>Week 5 Introduction to cue-controlled relaxation, partial relaxation and applied relaxation (integration of brief relaxation techniques into daily activities).† Solving problems encountered in participant's</p>	14 Behaviour regulation	Self-monitoring	Week 1 Call: (Education on headache characteristics, how to identify & avoid)				
	11.Env context & resources –	Person x env interaction	Week 2: Monitoring stress				
	1. Knowledge	Procedural knowledge	Week 2: Monitoring headache related stress				
	2. Skills	Skills development	Week 2 Call: "Introduction to				
	1. Knowledge	Procedural knowledge	Week 3 Call: Practice relaxation, PMR and RI"				
	2. Skills	Skills development	Week 3 Call: Application of relaxation skills				
	1 Knowledge	Procedural knowledge	Week 4 Call: "Introduction to", deep breathing, imagery relaxation)				
	2 Skills	Skills development	Week 5 Call: "Introduction to"				

	<p>efforts to apply relaxation skills in preventing, aborting or coping with migraines. Thermal biofeedback training introduced. Practice biofeedback for 10 to 15 minutes per day.</p> <p>Week 6 Stress management training (evaluation of stressful events including headaches; identification of and challenging “stress-generating” thoughts and beliefs, preparing for stressful events, problem solving stressors as appropriate).‡ Solving problems in the application of relaxation, hand-warming and/or stress management skills to preventing, aborting and/or coping with headache</p> <p>Week 7 Introduction to activity “overload” and importance of activity pacing; information on use of palliative treatments (heat, cold, pressure, massage); use of relaxation and stress management skills for coping with migraines that do occur.</p> <p>Week 8 Identification of the most effective headache management skills for individual adolescent. Relapse prevention: coping with temporary setbacks and setting realistic</p>	<p>4. Beliefs about capabilities</p> <p>6. Beliefs about consequences</p> <p>13 Emotion</p> <p>1.Knowledge</p> <p>2. Skills</p> <p>14. Behaviour Regulation</p>	<p>Perceived behavioural control</p> <p>Outcome expectancies</p> <p>Stress, Positive/negative affect</p> <p>Procedural knowledge</p> <p>Skills development</p> <p>Action planning</p>	<p>Week 5 Practice biofeedback</p> <p>Week 6 Call: Negative thoughts</p> <p>Catastrophising</p> <p>Week 6 Call: Challenge thoughts and beliefs</p> <p>Week 6 Call: Preparing for stressful events</p> <p>Week 7 Call: “Introduction to” - activity pacing,</p> <p>Use of skills: relaxation and stress management skills</p> <p>Week 8 Call: Relapse prevention</p>					
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	expectations for long term migraine management.	12. Social Influence	Social support	Weekly calls					
Hicks (2006)	<p>Each participant in the treatment group also received a personalized relaxation tape, which included many relaxation and imagery techniques. A thought journal was also included in the package, to be used in conjunction with cognitive restructuring strategies.</p> <p>Participants were assigned skills to practice during the week (e.g., deep breathing) and these skills were then subsequently discussed in an e-mail or a telephone call.</p> <p>Over the 7-week treatment, the researcher regularly e-mailed and telephoned participants to check on progress and to review materials. During treatment, participants were contacted, according to a set schedule, by e-mail in weeks 1, 2, 3, 5, and 7 and by telephone in weeks 2, 4, and 6.</p> <p>Welcome Introduction to the program and therapist Responsibilities of therapist, child, and parent</p> <p>Session 1 Confidentiality and its limits Tracking pain with a diary Identifying pain triggers Overview of pain reduction methods</p>	<p>10. Memory attention and decision processes</p> <p>11.Env context & resources –</p> <p>2.Skills</p> <p>12. Social Influence</p> <p>14. Behaviour Regulation</p> <p>1.Knowledge</p>	<p>Attention</p> <p>Material resources</p> <p>Skills development</p> <p>Social support</p> <p>Self-monitoring</p>	<p>Attention focused through intervention Sessions 1-7: engage with treatment, decision making, processing info</p> <p>Personalised relaxation tape, thought journal</p> <p>Assigned skills to practice</p> <p>Weekly calls, regular emails</p> <p>Session 1: track pain using pain diary, identify pain triggers</p>		<p>Waitlist control</p> <p>Assigned skills and monitored via weekly calls</p>			

			Knowledge	Session 1: Information: overview of pain reduction methods				
	Session 2 Identifying pain management strategies already used Information on headaches and stomach aches (i.e., RAP) Setting goals for the program Deep breathing (included breathing exercise)	1.Knowledge	Knowledge	Session 2: Information: on headache and stomach ache				
		9 Goals	Goal setting	Session 2: goal setting				
		2 Skills	Skills development	Session 2: deep breathing skills				
	Session 3 Physical pain management methods (e.g., heat, cold, massage)	1 Knowledge	Procedural knowledge	Session 3: (Instruction on physical pain management methods)				
	Session 4 Effects of tension Benefits of relaxation Introduction to full body relaxation and imagery	1. Knowledge	Knowledge	Session 4: (Information on effects and benefits)				
	Session 5 Positive versus negative thinking Challenging negative thoughts			Session 4 "Introduction to" full body/imagery relaxation				

	<p>Problematic ways of thinking (e.g., catastrophizing) Strategies for changing thinking (e.g., thought stopping)</p> <p>Session 6 Benefits of social and physical activity The story of the tortoise and the hare— pacing yourself Mini relaxation</p> <p>Session 7 Planning to manage pain episodes Managing pain at school Recognizing progress Maintaining the program Check-ups</p>	<p>4. Beliefs about capabilities</p> <p>6. Beliefs about consequences</p> <p>1 Knowledge</p> <p>2. Skills</p> <p>9.Goals</p> <p>14 Behaviour Regulation</p>	<p>Perceived behavioural control</p> <p>Outcome expectancies</p> <p>Knowledge</p> <p>Practice</p> <p>Goal setting</p> <p>Action Planning</p>	<p>Session 5: Instruction on thought changing, challenging etc.</p> <p>Catastrophising</p> <p>Session 6: Information on benefits</p> <p>Session 6: Encouraged to practice mini relaxation</p> <p>Session 7 Planning to manage pain</p> <p>Session 7 Maintaining the program</p>				
Law (2015)	<p>Physical therapy</p> <p>Internet CBT adjunctive to specialized headache treatment</p> <p>Internet CBT - Psychological therapy included face-to-face cognitive behavioural therapy for pain management and/or biofeedback. In</p>	<p>10. Memory attention and decision processes</p> <p>1 Knowledge</p>	<p>Attention</p> <p>Knowledge</p>	<p>Attention focused through intervention</p> <p>Pain education: Face to face instruction on medication</p>	<p>Specialized headache treatment (medication management and physical therapy)</p> <p>Participants received one or more of the following interventions as recommended by their providers at the headache clinic: medication</p>	<p>10. Memory attention and decision processes</p> <p>1 Knowledge</p>	<p>Attention</p> <p>Knowledge</p>	<p>Attention focused through treatment</p> <p>Face to face instruction on medication</p>

	<p>addition, participants received access to an Internet CBT program (Web-based Management of Adolescent Pain; Web-MAP).</p> <p>Adolescents and parents were asked to complete one module per week over the course of 8 weeks, which were designed to be about 30 minutes in length. The program is travel themed.</p> <p>Adolescents completed modules on pain education and goal setting, relaxation training, distraction strategies, cognitive strategies, sleep and lifestyle, interventions, and relapse prevention and maintenance.</p>	<p>2. Skill</p> <p>1.Knowledge</p> <p>9 Goals</p> <p>2.Skills</p> <p>2.Skills</p>	<p>Skills development</p> <p>Knowledge</p> <p>Goal setting</p> <p>Skills development</p> <p>Skills development</p>	<p>management, physical therapy and/or psychological therapy</p> <p>Education module – Information</p> <p>Relaxation Module – relaxation skills training</p> <p>Pain education</p> <p>Sessions 1-8: Goal setting</p> <p>Module - distraction skills training</p> <p>Cognitive Restructuring Module – cognitive skills training</p>	<p>management, psychological therapy, and physical therapy.</p> <p>Psychological therapy included face-to-face cognitive behavioral therapy for pain management and/or biofeedback.</p>	<p>2. Skill</p> <p>12 Social Influences</p>	<p>Skills development</p> <p>Social support</p>	<p>management, physical therapy and/or psychological therapy</p> <p>Face to face support</p>
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	<p>The program is interactive and required adolescents and parents to respond to queries which then provided tailored and personalized instruction and assignments.</p> <p>The program also includes videos of peer models and audio files of relaxation exercises.</p> <p>In six of the eight modules, parents and adolescents were given behavioural assignments focused on practice of skills taught in that module.</p> <p>Participants were instructed to work on the assignment for 1 week, and then to log back into the website to report on their progress with learning the skills in that module.</p> <p>These assignments were similar to weekly assignments used in face-to-face CBT for pain management. Assignment completion was required before participants were allowed to move on to the next module.</p>	<p>4. Beliefs about capabilities</p> <p>6. Beliefs about consequences</p> <p>11. Env context & resources</p> <p>12. Social Influence</p> <p>2. Skills</p> <p>1 Knowledge</p> <p>10. Memory attention and decision processes</p> <p>12. Social Influence</p>	<p>Perceived behavioural control</p> <p>Outcome expectancies</p> <p>Resources</p> <p>Social norms</p> <p>Skills practice</p> <p>Knowledge</p> <p>Attention control</p> <p>Social support</p>	<p>Cognitive Restructuring Module - Cognitive strategies</p> <p>Modules 1-8: videos, audio files</p> <p>Peer models</p> <p>Modules 1-8 (instruction, encouragement / practice of skills)</p> <p>Session-based information on sleep and lifestyle</p> <p>Module: distraction strategies</p>					
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	The online coach provided asynchronous feedback on each assignment via an online message center. A manual was developed to guide therapist responses to assignments. Responses focused on review of progress, encouraging skills practice, and problem-solving barriers to implementing skills.	2 Skills	Practice	Regular feedback via message centre Online Coach encouraged skills practice				
McGrath (1992)	<p>The self-administered treatment program consisted of an 8 chapter treatment manual and cassette tapes (McGrath et al. 1990a). Each week a different chapter was assigned and the adolescents could either read the manual or listen to instructions by tape or both. Each chapter focused on different coping and relaxation strategies.</p> <p>Subjects in the self-administered treatment were seen for an initial appointment and contacted weekly by telephone to answer any questions and to discuss homework assignments received by mail.</p> <p>The assignments consisted of a headache diary, a coping exercise and a chapter comprehension questionnaire. [The program for the clinic group was identical to the self-administered group, except that instead of receiving the manual and tapes and being telephoned weekly, each was seen individually by a trained therapist.]</p>	<p>10. Memory attention and decision processes</p> <p>11. Env context & resources</p> <p>12. Social Influence</p> <p>14. Behaviour Regulation</p> <p>1 Knowledge</p>	<p>Attention</p> <p>Resources</p> <p>Social support</p> <p>Self-monitoring</p>	<p>Attention focused through intervention</p> <p>Treatment manual and tapes</p> <p>Weekly calls</p> <p>Headache diary</p>	<p>The control group subjects were given a list of common triggers that can cause migraines such as different foods, too much sun, and too much exercise. In an initial treatment session with a therapist, they were asked to become aware of triggers that caused them headaches and to avoid them.</p> <p>They were also taught to use a brainstorming technique to deal with stressful situations.</p> <p>The therapist contacted the subjects weekly by telephone to monitor their progress. The control group was designed as a credible placebo that would have a similar amount of therapist contact as the self-administered treatment.</p>	<p>1 Knowledge</p> <p>14. Behaviour Regulation</p> <p>2 Skills</p> <p>12 Social Influences</p>	<p>Knowledge</p> <p>Breaking habit</p> <p>Skills development</p> <p>Social Support</p>	<p>Lis of triggers</p> <p>Avoiding common triggers</p> <p>Face to face – taught brainstorming technique</p> <p>Weekly telephone calls</p>

	Week 1 Rationale and explanation of coping exercise and relaxation with tension		Knowledge	Weeks 1: Rationale				
	Week 2 Cognitive restructuring	4. Beliefs about capabilities	Perceived behavioural control	Week 2 and 3: Chapter used to teach thought challenging,				
	Week 3 Examining unrealistic beliefs and relaxation without tension	6. Beliefs about consequences	Outcome expectancies	examining unrealistic beliefs and relaxation				
	Week 4 Distraction strategies	2 Skills	Skills development	Week 4: Distraction strategies				
		10. Memory attention and decision processes	Attention control	Distraction strategies				
	Week 5 Imagery, behavior rehearsal, mental activities, relaxation with imagery	2.Skills	Skills development	Week 5: Imagery, relaxation, distraction strategies				
		2.Skills	Skills practice	Week 5: Chapter used to encourage rehearsal				
	Week 6 Assertiveness	2 Skills	Skills development	Week 6: Chapter used to teach				

	<p>Week 7 Problem solving</p> <p>Week 8 Summary of coping strategies</p>	<p>14. Behaviour Regulation</p> <p>2.Skills</p>	<p>Action planning</p> <p>Knowledge</p>	<p>assertiveness / communication skills</p> <p>Week 7: (Chapter used to teach problem solving)</p> <p>Week 8: Chapter used to review all tasks</p>				
Palermo (2016)	<p>Adolescents and parents in the Internet CBT condition received access to the full Web-MAP2 program including education about chronic pain, training in behavioral and cognitive coping skills instruction in increasing activity participation, and education about pain behaviors and parental operant and communication strategies using an engaging interactive format.</p> <p>Participants in the Internet CBT condition had access to 5 functional components of the Web program, (1) treatment modules,</p> <p>(2) assessments and daily diaries, (3) compass (audio files of relaxation strategies),</p> <p>4) passport (progress tracker), and</p>	<p>10. Memory attention and decision processes</p> <p>1.Knowledge</p> <p>14. Behaviour Regulation</p> <p>11.Env context & resources</p> <p>14. Behaviour Regulation</p>	<p>Attention</p> <p>Knowledge</p> <p>Self-monitoring</p> <p>Material resources</p> <p>Self-monitoring</p>	<p>Attention focused through intervention</p> <p>Pain education</p> <p>Daily diaries</p> <p>Audio files</p>	<p>The pain education control group served as an attention control condition, equalizing time, attention, and computer usage. The control version of the Web-MAP study Web site had 2 functional components: (1) modules with information compiled from publicly available educational Web sites about pediatric chronic pain management (eg, National Headache Foundation, etc), and (2) diary and assessments. The control Web site did not provide access to behavioral and cognitive skills training. Adolescents and parents were instructed to log onto the Web program weekly at the same interval as the CBT group to read information about pediatric chronic pain. Reminders to access the Internet program were provided by study staff every 2</p>	<p>10. Memory attention and decision processes</p> <p>1.Knowledge</p> <p>14. Behaviour Regulation</p>	<p>Attention</p> <p>Knowledge</p> <p>Self-monitoring</p>	<p>Attention control condition</p> <p>Information compiled</p> <p>Pain diary</p>

	<p>(5) cognitive skills (eg, reducing negative thoughts),</p> <p>(6) sleep hygiene and lifestyle,</p> <p>(7) staying active (eg, activity pacing, pleasant activity scheduling), and</p> <p>(8) relapse prevention.</p> <p>Vignettes, videos of peer models, illustrations, and reinforcing quizzes are used throughout the program to increase interactivity.</p> <p>At some destinations, adolescents receive online postcards from previous places</p>	<p>4. Beliefs about capabilities</p> <p>6. Beliefs about consequences</p> <p>1 Knowledge</p> <p>2 Skills</p> <p>1 Knowledge</p> <p>12. Social Influence</p> <p>11. Env context & resources</p> <p>10. Memory attention and</p>	<p>Action Planning</p> <p>Perceived behavioural control Outcome expectancies</p> <p>Knowledge</p> <p>Skills development</p> <p>Knowledge</p> <p>Social norms</p> <p>Resources / material resources</p>	<p>Module 4: Implementation coping plans / action planning</p> <p>Module 5: Instruction on thought challenging</p> <p>Module 6: Information on sleep & lifestyle</p> <p>Module 7: Instruction on activity pacing and scheduling)</p> <p>Module 8: Information on relapse prevention</p> <p>Peer models</p> <p>Modules 1-8: (audio and video files, quizzes)</p>					
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	<p>they have visited reminding them to practice skills. Adolescents and parents interact with the program by identifying personal goals and entering information, which allowed tailoring and personalization of information for weekly behavioral assignments. Adolescents and parents were asked to complete 1 module per week, designed to be analogous to weekly, clinician-delivered in-person CBT. Participants (youth and parents) spent time practicing skills and completing assignments in 6 of the 8 modules.</p> <p>A message center allows communication between an online study coach and participant about each assignment. Adolescents and parents could also initiate messages to the coach at any time during the treatment period. Assignments were reviewed by 5 study coaches</p> <p>Coaches responded to each message sent by participants. Responses included praise for skills practice (eg, “Nice job practicing guided imagery!”), strategies to overcome barriers to using skills (eg, “Try practicing guided imagery at the same time every day”), and content to build rapport (eg, “Did you do anything fun over the weekend?”).</p>	<p>decision processes</p> <p>12. Social Influence</p> <p>7.Reinforcement</p>	<p>Memory</p> <p>Social support</p> <p>Rewards</p>	<p>Postcards</p> <p>Coach</p> <p>Verbal praise</p>				
<p>Palermo (2009)</p>	<p>Participants in the Internet treatment group also continued with their medical care recommended by their subspecialty physician (involving on average 0–2 specialty care visits), and were asked not</p>	<p>10. Memory attention and decision processes</p>	<p>Attention</p>	<p>Attention focused through intervention</p>	<p>Participants in the wait-list control group continued with their medical care recommended by their subspecialty physician, which for most patients involved 0–2 visits at the specialty clinic</p>	<p>10. Memory attention and decision processes</p>	<p>Attention</p>	<p>Attention focused through website access</p> <p>Pain diary assessment</p>

<p>to initiate psychotherapy during the 8-week treatment period.</p> <p>Web- MAP has three main sections: passport (home) page, treatment modules, and daily diary. Children and parents were each asked to log on once per week to the website to complete a module and assignment, designed to take about 30 min to complete.</p> <p>The program was travel-themed, and children and parents visited eight modules in destinations (e.g., China, Costa Rica; 1 per week), each focused on a different skill/topic. Children and parents interacted with the web program through completing fillable responses to queries (e.g., listing current stressors), which then tailored and personalized instructions and assignments. There were also several tasks that children completed using animations on the program (e.g., packing a suitcase to learn tips on relaxation).</p> <p>Web-MAP contains over 200 page views and makes use of multimedia elements to enhance delivery of information such as video files of teens who have experienced chronic pain and their parents, and audio files of deep breathing and muscle relaxation instructions.</p> <p>Each treatment module served as an online analog for the weekly sessions typically used when delivering CBT in a</p>	<p>14. Behaviour Regulation</p> <p>12. Social Influence</p> <p>11.Env context & resources</p>	<p>Self-monitoring</p> <p>Social norms</p> <p>Material resources</p>	<p>Modules 1-8 Encouragement to use daily diaries</p> <p>Teens</p> <p>Modules 1-8: audio and video files</p>	<p>over the treatment period. Children received access to the website to complete diary assessments at pre- and post-treatment assessments. Participants were asked not to initiate psychotherapy during the 8-week wait period.</p>	<p>14 Behaviour Regulation</p>	<p>Self-monitoring</p>	
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	<p>6) sleep hygiene and lifestyle,</p> <p>(7) staying active,</p> <p>(8) relapse prevention.</p> <p>The eight parent modules included: (1) education about chronic pain, (2) recognizing stress and negative emotions, (3) operant strategies I, (4) operant strategies II, (5) modeling, (6) sleep hygiene and lifestyle, (7) communication, and (8) relapse prevention.</p> <p>The child modules included instruction in identifying stress, applying deep breathing and progressive muscle relaxation, and modifying cognitions about pain and functional ability. In addition, one lesson in the child program focused on enhancing children's sleep habits (instruction in adequate sleep duration and sleep habits) and increasing their physical activity participation through goal setting and activity pacing.</p>	<p>1 Knowledge</p> <p>2 Skills</p> <p>1 Knowledge</p>	<p>Outcome expectancies</p> <p>Knowledge</p> <p>Skills development</p> <p>Knowledge</p>	<p>Module 6: Information on sleep & lifestyle</p> <p>Module 7: Instruction on being active</p> <p>Module 8: Information on relapse prevention</p>					
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<p>Rapoff (2014)</p>	<p>Participants in the Headstrong Group received the Headstrong CD-ROM program (see Table 1) while continuing to follow their treating neurologist's recommendations and prescriptions. Children were asked to complete the program in 4 weeks, with approximately 1 lesson per day and were required to take simple quizzes to assess their processing of the information presented.</p> <p>As with the Control group, various passwords and homework assignments were embedded within the program to ensure that children were adherent in viewing and applying the material.</p> <p>The layout of the cognitive-behavioral component of the CD-ROM intervention was similar to the education component of the Control group (including graphics, audio narration, music, clickable progress controls, passwords, homework assignments). However, the treatment component also contained lessons on how to use various empirically supported cognitive-behavioral treatments to self-manage recurrent headaches. Specifically, week two focused on relaxation methods (including a rationale with narrated and illustrated instructions on guided imagery, deep breathing, and progressive muscle relaxation), week three focused on problem-solving and stress management, and week four targeted pain behavior and parental response to pain as well as a review of the previous weeks' lessons. A workbook accompanied the Headstrong</p>	<p>10. Memory attention and decision processes</p> <p>11.Env context & resources</p>	<p>Attention</p> <p>Material resources</p>	<p>Attention focused through intervention</p> <p>CD-ROMs, record sheets, relaxation logs</p>	<p>Children in the control group continued to follow the recommendations and prescriptions of their treating neurologist. Typically, treatment included acute medications (e.g., NSAIDs, triptans, and muscle relaxants) and/ or preventative medications for children with a frequency of headache greater than one per week (e.g., anticonvulsants, antidepressants, beta blockers).</p> <p>Control participants received a developmentally appropriate educational CD-ROM program (see Table 1) containing information about primary headaches (i.e., types of primary headache, how headaches are assessed, typical symptoms, typical triggers, prevalence, etiology, and the multiple components of pain). The information contained in the education CD-ROM was more in-depth but similar to that contained in the first part of the Headstrong program (i.e., Module #1: Education). The education CD-ROM also covered health habits (e.g., sleep, diet, physical activity), but no "active" psychological headache therapies (e.g., relaxation, cognitive restructuring) were contained in this program.</p>	<p>11.Env context & resources</p> <p>1 Knowledge</p> <p>1 Knowledge</p>	<p>Material resources</p> <p>Procedural knowledge</p> <p>Knowledge</p>	<p>Educational CD-ROM</p> <p>Face to face recommendations of neurologist</p> <p>Educational CD-ROM</p>
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	<p>CD-ROM and contained all the supplementary material required for the self-management intervention.</p> <p>Weekly phone calls continued during this intervention phase to answer questions about the CD-ROMs and to remind participants to complete and return password sheets.</p> <p>Week 1 Headache education and cognitive behavioural model of pain</p> <ol style="list-style-type: none"> 1. Introduction 2. Types of headache 3. Prevalence of headache 4. Features of headache 5. How headache is diagnosed 6. The pain puzzle 7. Headache triggers <p>Week 2 Relaxation</p> <ol style="list-style-type: none"> 1. Rationale for relaxation 2. How to use guided imagery 3. How to use deep breathing 4. How to use progressive muscle relaxation <p>Week 3 Cognitive restructuring</p> <ol style="list-style-type: none"> 1. Rationale for coping 2. Thought changing 3. Problem solving 	<p>12. Social Influence</p> <p>1. Knowledge</p> <p>1. Knowledge</p> <p>1 Skills</p> <p>1. Knowledge</p> <p>4. Beliefs about capabilities</p>	<p>Social Support</p> <p>Knowledge</p> <p>Knowledge</p> <p>Skills development</p> <p>Knowledge</p>	<p>Weekly calls</p> <p>Week 1 CD-ROM: Pain education</p> <p>Week 2 CD-ROM: Rationale for relaxation</p> <p>Week 2 CD-ROM Relaxation: How to..instructions</p> <p>Week 3 CD-ROM: Rationale for coping</p>	<p>Children were asked to complete the program in 4 weeks, with approximately 1 lesson per day. The control CD-ROM controlled for the amount of headache education the two groups received and the time taken to completing the program.</p> <p>Weekly phone calls continued during this intervention phase to answer questions about the CD-ROMs and to remind participants to complete and return password sheets.</p> <p>Week 1 Headache education</p> <ol style="list-style-type: none"> 1. Introduction 2. Types of headache 3. Prevalence of headache 4. Features of headache 5. How headache is diagnosed <p>Week 2 Cognitive-behavioural model of pain</p> <ol style="list-style-type: none"> 1. Introduction to the pain puzzle 2. Puzzle piece 1: Nociception 3. Puzzle piece 2: Thoughts <p>Week 3: Cognitive-behavioural model of pain</p> <ol style="list-style-type: none"> 1. Puzzle piece 3: Feelings 	<p>12. Social Influence</p> <p>10. Memory attention and decision processes</p> <p>1 Knowledge</p>	<p>Social support</p> <p>Memory</p> <p>Knowledge</p>	<p>Weekly calls</p> <p>Remind participants</p> <p>Weeks 1-4 CD-ROM Headache education</p>
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	<p>Week 4 Pain behaviours</p> <ol style="list-style-type: none"> 1. Positive and negative pain behaviours 2. Importance of keeping active 3. Review of all lessons 	<p>6. Beliefs about consequences</p> <p>14. Behaviour regulation</p> <p>1 Knowledge</p>	<p>Perceived behavioural control</p> <p>Outcome expectancies</p> <p>Action planning</p> <p>Knowledge</p>	<p>Week 3 CD-ROM: Cognitive restructuring, thought changing, problem-solving</p> <p>Week 4 CD-ROM Pain Behaviours: Instruction on coping behaviours</p> <p>Review</p>	<p>2. Puzzle piece 4: Behavior</p> <p>Week 4: Headache triggers</p> <ol style="list-style-type: none"> 1. Introduction to headache triggers 2. Key headache triggers: diet and sleep 			
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Stinson (2010)	<p>For the experimental group, the intervention was a 12-week multicomponent treatment protocol that consisted of self-management strategies, disease-specific information, and social support that was available in English and French. It was delivered on a restricted website and through regular telephone contact with a trained coach (a non-healthcare professional with an undergraduate degree in psychology).</p> <p>The 12 modules for adolescents included learning about the different types of arthritis, understanding how arthritis is diagnosed, arthritis medications, managing symptoms (pain, stiffness, and fatigue), managing stress and negative thoughts, relaxation, distraction, other types of care (exercise, nutrition, splints), self-monitoring and supports, lifestyle issues, and looking ahead (education, vocation, transitional care issues). There were 2 modules specifically for parents/caregivers, to help them encourage healthy behavior (the effect of arthritis and helping parents learn to let go). Parents were able to view the materials on the teen website. The content is multi-layered, interactive,</p>	<p>10. Memory attention and decision processes</p> <p>12. Social Influence</p>	<p>Attention</p> <p>Social support</p>	<p>Attention focused through intervention</p> <p>Regular telephone contact from a trained coach</p>	<p>For the attention control group, credible attention strategies consisting of weekly phone contact by a trained research assistant to discuss adolescents' "own best efforts" at managing their JIA were administered over the 12-week period. The research assistant used a standardized script for these calls. Participants received a mean of 1.4 phone calls per week to maintain weekly contact; the average duration of calls was 3 min (range 2–6 min). If participants asked for information related to self-management of their JIA, the research assistant responded that she was not trained to provide such information and redirected them to their doctor or nurse.</p>	<p>10. Memory attention and decision processes</p> <p>12. Social Influence</p>	<p>Attention</p> <p>Social support</p>	<p>Attention control</p> <p>Weekly call</p>

	<p>written at a grade 6-7 level and geared to the self-management needs identified by adolescents and their parents.</p> <p>Adolescents were asked to log on to the site once per week to complete a module that was designed to take between 20 and 30 minutes. At the beginning of the program, adolescents developed their own personal goals for the program and kept track of their progress in "My Journal."</p> <p>The intervention group was contacted by a coach on a weekly basis for 12 weeks using standardized scripts. The weekly telephone calls were structured and the primary coach duties were to (1) review the previous week's homework, knowledge quiz, and goals; (2) determine whether the participant completed the module and answer questions regarding the material and/or practice exercises (self-management strategies); and (3) provide guidance and help solve any problems that had arisen. If participants asked questions that the coaches could not answer, the coach redirected them to their rheumatology provider. The coach also monitored discussion board postings daily. All calls were recorded to ensure integrity of the intervention.</p> <p>Participants received a mean of 1.6 phone calls per week to maintain contact, with the average duration of calls 17.3 min (range 7–30 min).</p>	<p>14 Behaviour Regulation</p> <p>9 Goals</p> <p>1.Knowledge</p> <p>1.Knowledge</p> <p>2.Skills</p> <p>13 Emotion</p> <p>2.Skills</p> <p>1.Knowledge</p>	<p>Self-monitoring</p> <p>Goals setting</p> <p>Knowledge</p> <p>Knowledge</p> <p>Skills development</p> <p>Stress</p> <p>Skills development</p>	<p>My Journal used to track progress</p> <p>Online facility to set own goals</p> <p>Online, Session 1, Understanding Arthritis</p> <p>Online Session 2 Understanding diagnoses</p> <p>Online, Session 3 Managing symptoms</p> <p>Online Session 4, Instruction on managing stress</p> <p>Online Session 5, Instruction on relaxation skills</p>					
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	<p>Session 1: About Arthritis Session 2: Understanding Diagnosis Session 3: Managing Your Symptoms Session 4: Managing Stress Session 5: Relaxation Session 6: Arthritis Medications Session 7: Distraction Session 8: Other Types of Care Session 9: Managing Your Thoughts Session 10: Therapies, Self-monitoring, and Supports Session 11: Your Lifestyle Session 12: Looking Ahead</p>	<p>2.Skills</p> <p>10. Memory attention and decision processes</p> <p>1.Knowledge</p> <p>4. Beliefs about capabilities</p> <p>6. Beliefs about consequences</p> <p>14 Behaviour Regulation</p> <p>1 Knowledge</p>	<p>Knowledge</p> <p>Skills development</p> <p>Attention control</p> <p>Knowledge</p> <p>Perceived behavioural control Outcome expectancies</p> <p>Self-monitoring</p> <p>Knowledge</p>	<p>Online, Session 6 Educational materials on medication management</p> <p>Online Session 7, Instruction on distraction</p> <p>Online Session 7, Distraction strategies</p> <p>Online, Session 8 Educational materials on types of care etc.</p> <p>Online, Session 9 Instruction on how to manage your thoughts</p> <p>Online, Session 10 Self-monitoring, My journal</p> <p>Online, Session 11 Educational materials on</p>					
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		1 Knowledge	Knowledge	lifestyle/self-care etc. Online, Session 12 Information on future planning				
Trautmann (2010)	<p>In the first training module all participants received information about mechanisms, symptoms and types of headache and the role of stress as a trigger of attacks. All self-help modules for the conditions were accessed on the Internet using passwords. The three groups underwent six weeks of the training program with six modules weekly, including homework exercises and e-mail contact to discuss the week's module topics with their therapists.</p> <p>CBT CBT was adapted from the manualized face-to-face group therapy program devised by Denecke and Kro"ner-Herwig (2000) for children with recurrent headache. CBT was reduced from 8 to 6 sessions in a self-help format, and the protocol was adapted to adolescents up to 18 years. While the first module presented education on headaches, the second unit focused on stress management (perception of own stress symptoms, coping with stress).</p>	<p>10. Memory attention and decision processes</p> <p>1. Knowledge</p> <p>13 Emotion</p>	<p>Attention</p> <p>Knowledge</p> <p>Stress,</p>	<p>Attention focused through intervention</p> <p>Module 1 Education on headache</p> <p>Module 2 Instruction on stress management</p>	<p>In the first training module all participants received information about mechanisms, symptoms and types of headache and the role of stress as a trigger of attacks. All self-help modules for the conditions were accessed on the Internet using passwords. The three groups underwent six weeks of the training program with six modules weekly, including homework exercises and e-mail contact to discuss the week's module topics with their therapists.</p> <p>EDU Participants in the EDU group received only the first self-help module (education on headache), but they had the same number of e-mail contacts as those in the CBT and AR. The e-mails focused on the diary records of the previous week (e.g. Did you have any headache last week? What did you do?), rather than on cognitive behavioural elements or applied relaxation</p>	<p>10. Memory attention and decision processes</p> <p>1 Knowledge</p> <p>12. Social Influence</p>	<p>Attention</p> <p>Knowledge</p> <p>Social support</p>	<p>Attention focused through intervention</p> <p>Module 1 Education on headache pain</p> <p>Regular emails</p>

	<p>In the following modules the participants acquired progressive relaxation techniques, cognitive restructuring (identification of dysfunctional cognitions regarding headache and stress and identifying functional cognitions), self-assurance strategies (being pro-active and sensitive to one's own needs), as well as problem solving.</p> <p>Participants of the CBT were offered a CD with relaxation instructions (a full relaxation protocol involving tensing and relaxing of major muscle groups, beginning with the upper body and proceeding to the lower body), and they could download the relaxation instructions from the training website.</p> <p>In CBT and AR, each e-mail from the participants responded to the assigned exercises and reported on their headache in the previous week</p> <p>In addition, participants received a list of questions related to the weekly training module to demonstrate their understanding of the learning targets. The e-mail contents were documented and recorded by the therapists and randomly checked by the first author. Two additional e-mail contacts (booster) were performed at week 4 and 8 after the end of training program. The participants were reminded of the coping strategies learned during the training and advised to continue practicing them in their daily lives.</p>	<p>2.Skills</p> <p>4. Beliefs about capabilities</p> <p>6. Beliefs about consequences</p> <p>11.Env context & resources</p> <p>12. Social Influence</p> <p>1 Knowledge</p> <p>2 Skills</p>	<p>Skills development</p> <p>Perceived behavioural control</p> <p>Outcome expectancies</p> <p>Material resources</p> <p>Social support</p> <p>Procedural knowledge</p> <p>Practice</p>	<p>Module Instruction on relaxation skills</p> <p>Module: Instruction on thought challenging</p> <p>Module Instruction on problem solving</p> <p>CD with instructions</p> <p>Email correspondence</p> <p>Emailed - List of question functioned as review of skills</p> <p>Email reminders to continue practice</p>	<p>instructions. This condition served as an active control group.</p> <p>Participants in the EDU exclusively reviewed their experiences of headache (self-monitoring in diaries) in e-mails.</p> <p>In addition, participants received a list of questions related to the weekly training module to demonstrate their understanding of the learning targets. The e-mail contents were documented and recorded by the therapists and randomly checked by the first author. Two additional e-mail contacts (booster) were performed at week 4 and 8 after the end of training program. The participants were reminded of the coping strategies learned during the training and advised to continue practicing them in their daily lives.</p> <p>They received the "modules" on a weekly basis and were encouraged to download and read the text material and to print out the information and exercises for practice. Passwords were given by e-mail so that they could access the following week's training.</p> <p>If participants were unable to connect to the Internet, they had</p>	<p>14 Behaviour Regulation</p> <p>1 Knowledge</p>	<p>Self-monitoring</p> <p>Procedural knowledge</p>	<p>Diaries used to self-monitor</p> <p>Emailed - List of question functioned as review of skills</p>
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	<p>They received the “modules” on a weekly basis and were encouraged to download and read the text material and to print out the information and exercises for practice. Passwords were given by e-mail so that they could access the following week’s training.</p> <p>If participants were unable to connect to the Internet, they had the option to contact a therapist by phone. Responses were provided within 48 h. In the treatment phase, therapists encouraged the participants to follow the scheduled program by reading the information. If participants did not answer, they received a friendly reminder from the therapist.</p> <p>AR AR follows the training developed by Ost (1987). The self-help modules contained only several phases from the original training (Ost, 1987): progressive relaxation, cue-controlled relaxation and differential relaxation. Participants were offered a CD with these specific instruction tracks for the different stages of AR training to be used at home (4 tracks: a full relaxation protocol common to the CBT CD, one track of cue-controlled relaxation, two tracks for differential relaxation).</p> <p>In CBT and AR, each e-mail from the participants responded to the assigned</p>	<p>10. Memory attention and decision processes</p> <p>10. Memory attention and decision processes</p> <p>2 Skills</p> <p>11.Env context & resources</p>	<p>Memory</p> <p>Attention</p> <p>Skills development</p> <p>Material resources</p>	<p>Reminders</p> <p>Attention focused through intervention</p> <p>Instruction on relaxation training</p> <p>CD instructions</p>	<p>the option to contact a therapist by phone. Responses were provided within 48 h. In the treatment phase, therapists encouraged the participants to follow the scheduled program by reading the information. If participants did not answer, they received a friendly reminder from the therapist.</p>			
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	<p>exercises and reported on their headache in the previous week</p> <p>In addition, participants received a list of questions related to the weekly training module to demonstrate their understanding of the learning targets. The e-mail contents were documented and recorded by the therapists and randomly checked by the first author. Two additional e-mail contacts (booster) were performed at week 4 and 8 after the end of training program. The participants were reminded of the coping strategies learned during the training and advised to continue practicing them in their daily lives.</p> <p>They received the “modules” on a weekly basis and were encouraged to download and read the text material and to print out the information and exercises for practice. Passwords were given by e-mail so that they could access the following week’s training.</p> <p>If participants were unable to connect to the Internet, they had the option to contact a therapist by phone. Responses were provided within 48 h. In the treatment phase, therapists encouraged the participants to follow the scheduled program by reading the information. If participants did not answer, they received a friendly reminder from the therapist.</p>	<p>12. Social Influence</p> <p>10. Memory attention and decision processes</p> <p>2 Skills</p>	<p>Social support</p> <p>Memory</p> <p>Practice</p>	<p>Regular emails</p> <p>Reminders</p> <p>Emails - Encouraged to continue practicing</p>					
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Appendix 8: BCT Coding Sheet

‘0’: BCT is absent, or ‘1’: Present in all probability but the evidence is unclear, or ‘2’: Present beyond all reasonable doubt. .

	Connelly 2006		Cottrell 2007		Hicks 2006		Law 2015		McGrath 1992	
	CBT	Attention control	CBT	Triptan treatment	CBT	WLC	CBT	Active control	CBT	Attention control
1. Goals and planning										
1.1. Goal setting (behavior)	0	0	0	0	1	0	1	0	0	0
1.2. Problem solving	2	0	2	0	2	0	1	0	1	0
1.3. Goal setting (outcome)	0	0	0	0	0	0	0	0	0	0
1.4. Action planning	2	0	0	0	1	0	0	0	0	0
1.5. Review behavior goal(s)	0	0	0	0	0	0	0	0	0	0
1.6. Discrepancy between current behavior and goal	0	0	0	0	0	0	0	0	0	0
1.7. Review outcome goal(s)	0	0	0	0	0	0	0	0	0	0
1.8. Behavioral contract	0	0	0	0	0	0	0	0	0	0
1.9. Commitment	0	0	0	0	0	0	0	0	0	0
2. Feedback and monitoring										
2.1. Monitoring of behavior by others without feedback	0	0	0	0	0	0	0	0	0	0
2.2. Feedback on behaviour	0	0	1	0	2	0	1	0	0	0
2.3. Self-monitoring of behaviour	0	0	0	0	0	0	0	0	0	0
2.4. Self-monitoring of outcome(s) of behaviour	1	0	0	0	0	0	0	0	0	0

2.5. Monitoring of outcome(s) of behavior without feedback	0	0	0	0	0	0	0	0	0
2.6. Biofeedback	0	0	2	0	0	0	2	0	0
2.7. Feedback on outcome(s) of behavior	0	0	0	0	0	0	0	0	0
3. Social support									
3.1. Social support (unspecified)	0	0	1	0	2	0	2	2	2
3.2. Social support (practical)	0	0	1	0	0	0	0	0	0
3.3. Social support (emotional)	0	0	0	0	0	0	0	0	0
4. Shaping knowledge									
4.1. Instruction on how to perform the behavior	2	0	2	0	2	0	2	0	1
4.2. Information about Antecedents	0	0	0	0	1	0	0	1	0
4.3. Re-attribution	0	0	0	0	0	0	0	0	0
4.4. Behavioral experiments	0	0	0	0	0	0	0	0	0
5. Natural consequences									
5.1. Information about health consequences	0	0	0	0	0	0	0	0	0
5.2. Salience of consequences	0	0	0	0	0	0	0	0	0
5.3. Information about social and environmental consequences	0	0	0	0	2	0	0	0	0

5.4. Monitoring of emotional consequences	0	0	0	0	0	0	0	0	0
5.5. Anticipated regret	0	0	0	0	0	0	0	0	0
5.6. Information about emotional consequences	0	0	0	0	0	0	0	0	0
6. Comparison of behaviour									
6.1. Demonstration of the behavior	2	0	0	0	0	0	2	0	0
6.2. Social comparison	0	0	0	0	1	0	1	0	0
6.3. Information about others' approval	0	0	0	0	0	0	0	0	0
7. Associations									
7.1. Prompts/cues	0	0	0	0	1	0	0	0	0
7.2. Cue signalling reward	0	0	0	0	0	0	0	0	0
7.3. Reduce prompts/cues	0	0	0	0	0	0	0	0	0
7.4. Remove access to the reward	0	0	0	0	0	0	0	0	0
7.5. Remove aversive stimulus	0	0	0	0	0	0	0	0	0
7.6. Satiation	0	0	0	0	0	0	0	0	0
7.7. Exposure	0	0	0	0	0	0	0	0	0
7.8. Associative learning	0	0	0	0	0	0	0	0	0
8. Repetition and substitution									
8.1. Behavioral practice/rehearsal	0	0	2	0	2	0	2	0	1
8.2. Behavior substitution	0	0	0	0	0	0	0	0	0
8.3. Habit formation	0	0	0	0	0	0	0	0	0

8.4. Habit reversal	0	0	0	0	0	0	0	0	0
8.5. Overcorrection	0	0	0	0	0	0	0	0	0
8.6. Generalisation of target behavior	0	0	0	0	0	0	0	0	0
8.7. Graded tasks	0	0	0	0	0	0	0	0	0
9. Comparison of outcomes									
9.1. Credible source	0	0	1	1	0	0	0	0	0
9.2. Pros and cons	0	0	0	0	0	0	0	0	0
9.3. Comparative imagining of future outcomes	0	0	0	0	0	0	0	0	0
10. Reward and threat									
10.1. Material incentive (behavior)	0	0	0	0	0	0	0	0	0
10.2. Material reward (behavior)	0	0	0	0	0	0	0	0	0
10.3. Non-specific reward	0	0	0	0	0	0	0	0	0
10.4. Social reward	0	0	0	0	1	0	0	0	0
10.5. Social incentive	0	0	0	0	0	0	0	0	0
10.6. Non-specific incentive	0	0	0	0	0	0	0	0	0
10.7. Self-incentive	0	0	0	0	0	0	0	0	0
10.8. Incentive (outcome)	0	0	0	0	0	0	0	0	0
10.9. Self-reward	0	0	0	0	0	0	0	0	0
10.10. Reward (outcome)	0	0	0	0	0	0	0	0	0
10.11. Future punishment	0	0	0	0	0	0	0	0	0
11. Regulation									

11.1. Pharmacological support	2	2	2	1	0	0	0	2	0	0
11.2. Reduce negative emotions	0	0	2	0	0	0	0	0	0	0
11.3. Conserving mental resources	0	0	0	0	0	0	0	0	0	0
11.4. Paradoxical instructions	0	0	0	0	0	0	0	0	0	0
12. Antecedents										
12.1. Restructuring the physical environment	0	0	0	0	0	0	0	0	0	0
12.2. Restructuring the social environment	0	0	0	0	0	0	0	0	0	0
12.3. Avoidance/reducing exposure to cues for the behavior	0	0	0	0	0	0	0	0	0	0
12.4. Distraction	0	0	0	0	0	0	1	0	1	0
12.5. Adding objects to the environment	0	0	2	0	2	0	0	0	2	0
12.6. Body changes	2	0	2	0	2	0	2	0	2	0
13. Identity										
13.1. Identification of self as role model	0	0	0	0	0	0	0	0	0	0
13.2. Framing/reframing	2	0	0	0	1	0	1	0	2	0
13.3. Incompatible beliefs	0	0	0	0	0	0	0	0	0	0
13.4. Valued self-identify	0	0	0	0	0	0	0	0	0	0
13.5. Identity associated with changed behavior	0	0	0	0	0	0	0	0	0	0

14. Scheduled consequences									
14.1. Behavior cost	0	0	0	0	0	0	0	0	0
14.2. Punishment	0	0	0	0	0	0	0	0	0
14.3. Remove reward	0	0	0	0	0	0	0	0	0
14.4. Reward approximation	0	0	0	0	0	0	0	0	0
14.5. Rewarding completion	0	0	0	0	0	0	0	0	0
14.6. Situation-specific reward	0	0	0	0	0	0	0	0	0
14.7. Reward incompatible behavior	0	0	0	0	0	0	0	0	0
14.8. Reward alternative behavior	0	0	0	0	0	0	0	0	0
14.9. Reduce reward frequency	0	0	0	0	0	0	0	0	0
14.10. Remove punishment	0	0	0	0	0	0	0	0	0
15. Self-belief									
15.1. Verbal persuasion about capability	0	0	0	0	0	0	0	0	0
15.2. Mental rehearsal of successful performance	0	0	0	0	0	0	0	0	0
15.3. Focus on past success	0	0	0	0	0	0	0	0	0
15.4. Self-talk	0	0	0	0	2	0	0	0	0
16. Covert learning									
16.1. Imaginary punishment	0	0	0	0	0	0	0	0	0
16.2. Imaginary reward	0	0	0	0	0	0	0	0	0
16.3. Vicarious consequences	0	0	0	0	0	0	0	0	0

‘0’: BCT is absent, or ‘1’: Present in all probability but the evidence is unclear, or ‘2’: Present beyond all reasonable doubt.

	Palermo 2009		Palermo 2016		Rapoff 2014		Stinson 2010		Trautmann 2010		
	CBT	WLC	CBT	EDU Control	CBT	EDU Control	CBT	Attention control	CBT	AR	EDU Control
1. Goals and planning											
1.1. Goal setting (behavior)	0	0	2	0	0	0	0	0	0	0	0
1.2. Problem solving	2	0	0	0	1	0	0	0	2	0	0
1.3. Goal setting (outcome)	1	0	0	0	0	0	1	0	0	0	0
1.4. Action planning	0	0	1	0	0	0	0	0	0	0	0
1.5. Review behavior goal(s)	0	0	0	0	0	0	0	0	0	0	0
1.6. Discrepancy between current behavior and goal	0	0	0	0	0	0	0	0	0	0	0
1.7. Review outcome goal(s)	0	0	0	0	0	0	0	0	0	0	0
1.8. Behavioral contract	0	0	0	0	0	0	0	0	0	0	0
1.9. Commitment	0	0	0	0	0	0	0	0	0	0	0
2. Feedback and monitoring											
2.1. Monitoring of behavior by others without feedback	0	0	0	0	0	0	0	0	0	0	0
2.2. Feedback on behaviour	1	0	1	0	0	0	0	0	0	0	0
2.3. Self-monitoring of behaviour	0	0	1	0	0	0	0	0	0	0	0

2.4. Self-monitoring of outcome(s) of behaviour	0	0	0	0	0	0	0	0	0	0	0
2.5. Monitoring of outcome(s) of behavior without feedback	0	0	0	0	0	0	0	0	0	0	0
2.6. Biofeedback	0	0	0	0	0	0	0	0	0	0	0
2.7. Feedback on outcome(s) of behavior	0	0	0	0	0	0	0	0	0	0	0
3. Social support											
3.1. Social support (unspecified)	2	0	0	0	0	0	2	2	0	0	0
3.2. Social support (practical)	0	0	0	0	0	0	0	0	0	0	0
3.3. Social support (emotional)	0	0	1	0	0	0	0	0	0	0	0
4. Shaping knowledge											
4.1. Instruction on how to perform the behavior	2	0	2	0	2	0	1	0	2	2	0
4.2. Information about Antecedents	0	0	0	0	0	1	0	0	1	1	1
4.3. Re-attribution	0	0	0	0	0	0	0	0	0	0	0
4.4. Behavioral experiments	0	0	0	0	0	0	0	0	0	0	0
5. Natural consequences											
5.1. Information about health consequences	0	0	0	0	1	0	1	0	0	0	0
5.2. Salience of consequences	0	0	0	0	0	0	0	0	0	0	0
5.3. Information about social and environmental consequences	0	0	0	0	0	0	0	0	0	0	0

5.4. Monitoring of emotional consequences	0	0	0	0	0	0	0	0	0	0	0
5.5. Anticipated regret	0	0	0	0	0	0	0	0	0	0	0
5.6. Information about emotional consequences	0	0	0	0	0	0	0	0	0	0	0
6. Comparison of behaviour											
6.1. Demonstration of the behavior	2	0	2	0	1	0	0	0	0	0	0
6.2. Social comparison	0	0	1	0	0	0	0	0	0	0	0
6.3. Information about others' approval	0	0	0	0	0	0	0	0	0	0	0
7. Associations											
7.1. Prompts/cues	0	0	0	2	2	0	0	0	0	0	0
7.2. Cue signalling reward	0	0	0	0	0	0	0	0	0	0	0
7.3. Reduce prompts/cues	0	0	0	0	0	0	0	0	0	0	0
7.4. Remove access to the reward	0	0	0	0	0	0	0	0	0	0	0
7.5. Remove aversive stimulus	0	0	0	0	0	0	0	0	0	0	0
7.6. Satiation	0	0	0	0	0	0	0	0	0	0	0
7.7. Exposure	0	0	0	0	0	0	0	0	0	0	0
7.8. Associative learning	0	0	0	0	0	0	0	0	0	0	0
8. Repetition and substitution											
8.1. Behavioral practice/rehearsal	1	0	2	0	0	0	1	0	1	1	0
8.2. Behavior substitution	0	0	0	0	0	0	0	0	0	0	0
8.3. Habit formation	0	0	0	0	0	0	0	0	0	0	0

8.4. Habit reversal	0	0	0	0	0	0	0	0	0	0
8.5. Overcorrection	0	0	0	0	0	0	0	0	0	0
8.6. Generalisation of target behavior	0	0	0	0	0	0	0	0	0	0
8.7. Graded tasks	0	0	0	0	0	0	0	0	0	0
9. Comparison of outcomes										
9.1. Credible source	2	2	2	2	0	0	0	0	0	0
9.2. Pros and cons	0	0	0	0	0	0	0	0	0	0
9.3. Comparative imagining of future outcomes	0	0	0	0	0	0	0	0	0	0
10. Reward and threat										
10.1. Material incentive (behavior)	0	0	0	0	0	0	0	0	0	0
10.2. Material reward (behavior)	0	0	0	0	0	0	0	0	0	0
10.3. Non-specific reward	0	0	0	0	0	0	0	0	0	0
10.4. Social reward	2	0	2	0	0	0	0	0	0	0
10.5. Social incentive	0	0	0	0	0	0	0	0	0	0
10.6. Non-specific incentive	0	0	0	0	0	0	0	0	0	0
10.7. Self-incentive	0	0	0	0	0	0	0	0	0	0
10.8. Incentive (outcome)	0	0	0	0	0	0	0	0	0	0
10.9. Self-reward	0	0	0	0	0	0	0	0	0	0
10.10. Reward (outcome)	0	0	0	0	0	0	0	0	0	0
10.11. Future punishment	0	0	0	0	0	0	0	0	0	0

11. Regulation											
11.1. Pharmacological support	0	0	0	0	0	0	0	0	0	0	0
11.2. Reduce negative emotions	1	0	2	0	0	0	2	0	2	0	0
11.3. Conserving mental resources	0	0	0	0	0	0	0	0	0	0	0
11.4. Paradoxical instructions	0	0	0	0	0	0	0	0	0	0	0
12. Antecedents											
12.1. Restructuring the physical environment	0	0	0	0	0	0	0	0	0	0	0
12.2. Restructuring the social environment	0	0	0	0	0	0	0	0	0	0	0
12.3. Avoidance/reducing exposure to cues for the behavior	0	0	0	0	0	0	0	0	0	0	0
12.4. Distraction	0	0	0	0	0	0	2	0	0	0	0
12.5. Adding objects to the environment	0	0	0	0	2	2	0	0	2	2	0
12.6. Body changes	2	0	2	0	2	0	2	0	2	2	0
13. Identity											
13.1. Identification of self as role model	0	0	0	0	0	0	0	0	0	0	0
13.2. Framing/reframing	0	0	1	0	1	0	0	0	1	0	0
13.3. Incompatible beliefs	0	0	0	0	0	0	0	0	0	0	0
13.4. Valued self-identify	0	0	0	0	0	0	0	0	0	0	0
13.5. Identity associated with changed behavior	0	0	0	0	0	0	0	0	0	0	0

14. Scheduled consequences											
14.1. Behavior cost	0	0	0	0	0	0	0	0	0	0	0
14.2. Punishment	0	0	0	0	0	0	0	0	0	0	0
14.3. Remove reward	0	0	0	0	0	0	0	0	0	0	0
14.4. Reward approximation	0	0	0	0	0	0	0	0	0	0	0
14.5. Rewarding completion	0	0	0	0	0	0	0	0	0	0	0
14.6. Situation-specific reward	0	0	0	0	0	0	0	0	0	0	0
14.7. Reward incompatible behavior	0	0	0	0	0	0	0	0	0	0	0
14.8. Reward alternative behavior	0	0	0	0	0	0	0	0	0	0	0
14.9. Reduce reward frequency	0	0	0	0	0	0	0	0	0	0	0
14.10. Remove punishment	0	0	0	0	0	0	0	0	0	0	0
15. Self-belief											
15.1. Verbal persuasion about capability	0	0	0	0	0	0	0	0	0	0	0
15.2. Mental rehearsal of successful performance	0	0	0	0	0	0	0	0	0	0	0
15.3. Focus on past success	0	0	0	0	0	0	0	0	0	0	0
15.4. Self-talk	0	0	0	0	0	0	0	0	0	0	0
16. Covert learning											
16.1. Imaginary punishment	0	0	0	0	0	0	0	0	0	0	0
16.2. Imaginary reward	0	0	0	0	0	0	0	0	0	0	0
16.3. Vicarious consequences	0	0	0	0	0	0	0	0	0	0	0

Appendix 9: Characteristics of included studies and risk of bias tables

Connelly 2006

Bibliographic information	Connelly, M., Rapoff, M. A., Thompson, N., & Connelly, W. (2006) Headstrong: A pilot study of a CD-ROM intervention for recurrent pediatric headache. <i>Journal of Pediatric Psychology, 31</i> , 737–47.
Design of study (including randomisation procedure)	RCT. Two arms. Assessed at pre-treatment, post-treatment, two months, three months. <i>Randomisation procedure:</i> ‘Stratified (age) randomisation to one of two groups by a research assistant using a uniform random numbers table.’ (Pg. 740)
Demographics	<p>Participants (Pg.738): N = 37 Sex: 18F, 19M Mean age = 10.0, SD (1.66) Age range 7-12 years Diagnosis = Headache Pain duration (months) – not reported Attrition: 6/37</p> <p>Recruitment: Attended the outpatient neurology clinic at a large children’s hospital in the Midwestern part of the United States</p> <p>Recruitment period: Aug 2002 - Feb 2003. (Pg.738)</p> <p>Analyses/Sample: ‘Intent to treat’ (LOCF): N= 37</p> <p>‘As treated’: Start of treatment: n = 37 Post-treatment: n = 36 Follow-up: n = 31</p>
Intervention group	<p>“<i>Headstrong programme, CD-ROM</i>”</p> <p>Setting: Computer-based, at home Components: education, relaxation, thought-changing, problem solving and pain behaviour modification plus weekly telephone contact to address questions.</p> <p>Additional materials: None reported</p>

Comparison group	<p><i>“Wait-list condition”</i></p> <p>Standard care (following recommendations of neurologist) plus weekly telephone contact to encourage consistent record keeping (Pg. 740)</p> <p>Additional materials: Submit weekly record sheet (for assessment purposes)</p>
Outcomes	<p>Clinical significance: 50% or greater pre-post change on the Headache Index</p> <p>Pain Symptoms: Total Pain (Headache diary/index)</p> <p>Pain related disability outcome: Pediatric Migraine Disability Assessment (Ped-MIDAS)</p> <p>Depression outcome: none</p> <p>Anxiety outcome: none</p> <p>Quality of life outcome: none</p> <p>Treatment satisfaction outcome: not reported</p>

Risk of bias table

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Low risk	Randomly assigned to one of two groups by a research assistant using a uniform random numbers table
Allocation concealment (selection bias)	Low risk	Stratified by age (7–9 and 10–12), randomly assigned to one of two groups (treatment or wait-list control) by a research assistant using a Uniform Random Numbers (URN) table. No restriction was placed on the randomization such that unequal group sample sizes were possible.
Blinding of participants and personnel (performance bias)	Low risk	Study neurologists remained blind to randomisation condition Throughout the study. Chances of un-blinding were limited because follow-up appointments with the study neurologist were scheduled for 2 months following the initial assessment

Blinding of outcome assessment (detection bias)	Low risk	Measures completed at home and posted back
Incomplete outcome data (attrition bias)	Low risk	Attrition is described, but further description of completers and non-completers was not reported
Selective reporting (reporting bias)	Low risk	Data were fully reported
Other bias	Unclear risk	This research was supported in part by an educational grant from AstraZeneca LP.

Cottrell 2007

Bibliographic information	Cottrell, C, Drew, J, Gibson, J, Holroyd, K, & O'Donnell, F. (2007) Feasibility Assessment of Telephone-Administered Behavioral Treatment for Adolescent Migraine. <i>Headache: The Journal of Head and Face Pain</i> , 47(9), 1293–1302.
Design of study (including randomisation procedure)	RCT. 2 arms. Assessed at pre-treatment, post-treatment, and 8 months. <i>Randomisation procedure</i> : ‘Randomised, no description found in text.’
Demographics	Participants (Pg.3 & 11): N= 30 Sex: 15F, 15M Mean age = 14.1 (SD 1.91) Range = 12-17 years Diagnosis = Headache Pain duration (months) – not reported Recruitment: Neurologist referral and community Recruitment period not reported Analyses/Sample: ‘Intent to treat’ (LOCF): N= 30 ‘As treated’: Start of treatment: n = 30 Post-treatment: n = 30 Follow-up: n = 28
Intervention group	“ <i>Stop Migraines treatment</i> ”

	<p>Setting: Telephone-based, at home Components: education, goal-setting, relaxation training, problem solving, thermal biofeedback training, stress management training (thought-changing) and activity pacing.</p> <p>Additional materials: Manual, relaxation tapes, home biofeedback equipment.</p>
Comparison group	<p><i>“Triptan treatment”</i></p> <p>Triptan therapy (following recommendations of neurologist)</p>
Outcomes	<p>Clinical significance: 50% or greater pre-post change on the Headache Index Pain Symptoms: none Pain related disability outcome: hours disabled by pain Depression outcome: none Anxiety outcome: none Quality of life outcome: none Treatment satisfaction outcome: narrative synthesis</p>

Risk of bias table

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Unclear risk	“34 adolescents were randomized to treatment (16 TT and 18 TAT).” description of randomisation not provided
Allocation concealment (selection bias)	Unclear risk	No description found in text
Blinding of participants and personnel (performance bias)	Unclear risk	No description found in text
Blinding of outcome assessment (detection bias)	Unclear risk	No description found in text

Incomplete outcome data (attrition bias)	Unclear risk	Attrition completely reported but further description of completers and non-completers was not reported
Selective reporting (reporting bias)	High risk	Outcomes incompletely reported
Other bias	Unclear risk	Funding source: National Institutes of Health (NINDS #N32374) Declarations of interest: Dr. O'Donnell is an employee of OrthoNeuro Inc

Hicks 2006

Bibliographic information	Hicks, C. L., von Baeyer, C. L., & McGrath, P. J. (2006). Online psychological treatment for pediatric recurrent pain: A randomized evaluation. <i>Journal of Pediatric Psychology</i> , 31, 724–36.
Design of study (including randomisation procedure)	RCT. Two arms. Assessed at pre-treatment, 1-month post-treatment, 3 months. Randomisation procedure: ‘The 47 participants were stratified by age and pain severity and randomly assigned by blocks to either the treatment condition or the standard medical care wait-list condition.’
Demographics	Participants (Pg. 725): N= 47 Sex: 30F, 17M Mean age = 11.7 (range 9-16) Diagnosis = Headache and RAP Pain duration (months): 36 Recruitment: Community over a one-year period Analyses/Sample: ‘Intent to treat’ (LOCF): N= 47 ‘As treated’: Start of treatment: n = 47 Post-treatment: n = 37, Follow-up (one-month): n = 37, Follow-up (three-months): n = 32 Attrition: n = 15/47
Intervention group	“Internet CBT” (with internet and phone) Manual used: Yes Setting: Computer-based, at home

	Components: education, deep breathing, relaxation, imagery, cognitive strategies, healthy lifestyle choices.
Comparison group	Standard Care (Wait List)
Outcomes	<p>Clinical significance: 50% or greater pre-post change on summed total pain score.</p> <p>Pain outcome: Total Pain Score (pain diary: NRS intensity, frequency, index)</p> <p>Disability outcome: none</p> <p>Depression outcome: none</p> <p>Anxiety outcome: none</p> <p>Quality of life outcome: Pediatric Quality of Life Inventory (PEDSQL)</p> <p>Treatment satisfaction: narrative synthesis</p>

Risk of bias table

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Low risk	The 47 participants were stratified by age and pain severity and randomly assigned by blocks to either the treatment condition or the standard medical care wait-list condition.
Allocation concealment (selection bias)	Low risk	The 47 participants were stratified by age and pain severity and randomly assigned by blocks to either the treatment condition or the standard medical care wait-list condition.
Blinding of participants and personnel (performance bias)	Unclear risk	No description found in text
Blinding of outcome assessment (detection bias)	Low risk	Measures completed at home and submitted online
Incomplete outcome data (attrition bias)	Unclear risk	Attrition completely reported, but further description of completers and non-completers was not reported
Selective reporting (reporting bias)	Low risk	Data were fully reported
Other bias	Unclear risk	support received through the Peter Samuelson STARBRIGHT Foundation 2002 Dissertation Award in pediatric psychology and the Canadian Pain Society

		Small Grant for Local and Regional Initiatives. McGrath is supported by a Canada Research Chair.
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Law 2015

Bibliographic information	Law, E. F., Beals-Erickson, S. E., Noel, M., Claar, R., & Palermo, T. M. (2015). Pilot randomized controlled trial of internet-delivered cognitive-behavioral treatment for pediatric headache. <i>Headache</i> , 55(10), 1410–1425.
Design of study (including randomisation procedure)	Pilot RCT. Two arms. Assessed at pre-treatment, 8-10 weeks post-treatment, 3 months. Fixed, block randomisation (blocks of 10) using an online random number generator. Concealed random allocation (1:1 ratio), performed using an ID number in an excel spreadsheet, automatically performed using software (Web-MAP system)
Demographics	<p>Participants: N= 83 Sex: 68F, 15M Mean age = 14.5 (1.7) Diagnosis = Chronic headache Pain duration (months): NR</p> <p>Recruitment: Clinic, over a 2-year recruitment period.</p> <p>Analyses/Sample: ‘Intent to treat’ (LOCF): N= 83 ‘As treated’: 49 Start of treatment: n = 83 Post-treatment: n = 59 Follow-up (three-months): n = 49 Attrition: n = 34/83</p>
Intervention group	<p>“Internet CBT” (with internet and email) Setting: Internet-based, at home Components: The design and treatment content of Web-MAP was identical to the original version of the program. Components include education about chronic pain, recognising stress and negative emotions, deep breathing and relaxation, distraction, cognitive skills, sleep hygiene and lifestyle, staying active, relapse prevention.</p>

Comparison group	Specialized Headache Treatment - Participants received one or more of the following interventions as recommended by Headache their providers at the headache clinic: medication management, psychological therapy, and physical therapy.
Outcomes	<p>Clinical significance: 50% or greater pre-post change on summed total pain score.</p> <p>Pain outcome: Total Pain Score (pain diary: NRS intensity, frequency, index)</p> <p>Disability outcome: Activity Limitations Interview 21</p> <p>Depression outcome: Children’s Depression Inventory,</p> <p>Anxiety outcome: Revised Children’s Manifest Anxiety Scale.</p> <p>Treatment satisfaction: Treatment Evaluation Inventory short form</p>

Risk of bias table

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Low risk	Blocked randomization with blocks of 10 was used to assign participants to one of the two treatment conditions. An online number generator was used to produce the blocked randomization. Participants were allocated in a 1:1 ratio
Allocation concealment (selection bias)	Low risk	Group assignments were identified by ID number in an excel spreadsheet that was password protected and accessible only to a research coordinator who was blinded to participant recruitment, screening, and informed consent. Following completion of all pre-treatment assessments, the research coordinator accessed the excel spreadsheet to reveal the group assignment. This information was then programmed into the Web-MAP system, which generated a message on the website to each study participant revealing the instructions for their treatment assignment.

Blinding of participants and personnel (performance bias)	Unclear risk	Because of the nature of the intervention, it was not possible to blind participants or research staff to group status.
Blinding of outcome assessment (detection bias)	Low risk	Research coordinator who was blinded to group status conducted all assessment procedures that occurred in the clinic.
Incomplete outcome data (attrition bias)	Unclear risk	Attrition completely reported, but further description of completers and non-completers was not reported
Selective reporting (reporting bias)	Low risk	Data were fully reported
Other bias	Unclear risk	

McGrath 1992

Bibliographic information	McGrath, P. J., Humphreys, P., Keene, D., Goodman, J.T., Lascelles, M. A., Cunningham, S. J., et al. (1992) The efficacy and efficiency of a self-administered treatment for adolescent migraine. <i>Pain</i> , 49, 321–4.
Design of study (including randomisation procedure)	RCT. Three arms. Assessed at pre-treatment, post-treatment, three months and one-year follow-up. Randomisation procedure: no description found in text ‘Randomised to 1 of 8-week treatments’.
Demographics	Participants: N= 87 Sex: 63F, 24M Mean age = NR, range: 11-18 years Diagnosis = Migraine Pain duration (months): minimum of three months Recruitment: Clinician referral, recruitment period not reported. Analyses/Sample: ‘Intent to treat’ (LOCF): not reported ‘As treated’: 73 Start of treatment: n = 87 Post-treatment: n = 72 Follow-up (three and twelve-months): n = 72

	Attrition: n = 12/72
Intervention group	<p>“Telephone and manual-based CBT” (with manual, tapes and telephone) Setting: Home, manual and telephone Components: Each chapter focused on different coping and relaxation strategies. Rationale and explanation of coping exercise and relaxation with tension, cognitive restructuring, examining unrealistic beliefs and relaxation without tension, distraction strategies, imagery, behaviour rehearsal, mental activities, relaxation with imagery, assertiveness, problem solving and summary of coping strategies.</p>
Comparison group	Clinic group: identical to the self-administered group, except that instead of receiving the manual and tapes and being telephoned weekly, each was seen individually by a trained therapist.
Outcomes	<p>Clinical significance: 50% or greater pre-post change on summed total pain score. Pain outcome: Headache Index Disability outcome: none Depression outcome: Poznanski Depression Scale Anxiety outcome: none Treatment satisfaction: none</p>

Palermo 2009

Bibliographic information	Palermo, T. M., Wilson, A. C., Peters, M., Lewandowski, A., & Somhegyi, H. (2009) Randomized controlled trial of an internet delivered family cognitive behavioral therapy intervention for children and adolescents with chronic pain. <i>Pain, 146</i> (1-2), 205–13.
Design of study (including randomisation procedure)	RCT. Two arms. Assessed at pre-treatment and post-treatment. Fixed, block randomisation (blocks of 10) using an online random number generator. Research assistant, performed using an ID number in sealed envelopes
Demographics	Participants: N= 48

	<p>Sex: 35F, 13M Mean age = 14.8 (2.0 SD), range: 11-17 years Diagnosis = Headache, abdominal pain or musculoskeletal pain Pain duration (months): 30 months</p> <p>Recruitment: Clinic, 2 year. recruitment period</p> <p>Analyses/Sample: ‘Intent to treat’ (LOCF): n= 48</p> <p>‘As treated’: Start of treatment: n = 48 Post-treatment: n = 44 Follow-up (three months): n = 23 Attrition: n = 2/48</p>
Intervention group	<p>“Internet-based CBT” (with email) Setting: Internet-based and email Components: Interactive, self-guided Internet intervention to deliver family cognitive-behavioural therapy</p>
Comparison group	<p>Waitlist control group</p>
Outcomes	<p>Clinical significance: 50% or greater pre-post change on summed total pain score. Pain outcome: Clinically significant reduction (>50% reduction), mean pain intensity NRS (averaged over 7 days) Disability outcome: Child Activity and Limitations Interview (CALI) Depression outcome: Revised Child Anxiety and Depression Scale (RCADS). Anxiety outcome: none Treatment satisfaction: Treatment Evaluation Inventory-Short Form</p>

Risk of bias table

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Low risk	<p>Page 206 A fixed allocation randomization scheme was used. Specifically, we used blocked randomization with blocks of 10 to assign participants to the two treatment</p>

		conditions during the course of randomization. An online random number generator was used to produce the blocked randomization. Group assignments were identified by ID number in sealed envelopes during the 24-month recruiting period. Following completion of all pre-treatment assessments, a research coordinator opened the sealed envelope to reveal the group assignment.
Allocation concealment (selection bias)	Low risk	Page 206 A fixed allocation randomization scheme was used. Specifically, we used blocked randomization with blocks of 10 to assign participants to the two treatment conditions during the course of randomization. An online random number generator was used to produce the blocked randomization. Group assignments were identified by ID number in sealed envelopes during the 24-month recruiting period. Following completion of all pre-treatment assessments, a research coordinator opened the sealed envelope to reveal the group assignment.
Blinding of participants and personnel (performance bias)	Unclear risk	No description found in text - probably not done
Blinding of outcome assessment (detection bias)	Low risk	Measures completed at home and submitted online or mailed back
Incomplete outcome data (attrition bias)	Low risk	Attrition completely reported;
Selective reporting (reporting bias)	Low risk	Data were fully reported
Other bias	Unclear risk	This research was funded by the National Institutes of Health/National Institute of Child Health and Human Development and by the Doernbecher Foundation.

Palermo 2016

Bibliographic information	Palermo, T.M., Law, E.F., Fales, J., Bromberg, M.H., Jessen Fiddick, T., Tai, G. Internet-delivered cognitive-behavioral
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	treatment for adolescents with chronic pain and their parents: a randomized controlled multicenter trial. <i>PAIN</i> 157 (2016) 174–185
Design of study (including randomisation procedure)	RCT. Two arms. Assessed at pre-treatment and post-treatment 6- and 12-month follow-up (Page 175). Fixed, block randomisation (blocks of 4) using a computer-generated randomization schedule. Concealed random allocation automatically performed using software (Web-MAP2 system).
Demographics	<p>Participants: N= 273 Sex: 257F, 15M Mean age = 14.7 (SD 1.6), range: 11-17 years Diagnosis = Headache, abdominal pain or musculoskeletal pain Pain duration (months): not reported</p> <p>Recruitment: Pain Clinic, 3.5 yr. recruitment period, Sep 2011 to April 2014 (Page 175).</p> <p>Analyses/Sample: ‘Intent to treat’ (LOCF): n= 269</p> <p>‘As treated’: 273 Start of treatment: n = 266 Post-treatment: n = 264 Follow-up (three months): n = 264 Attrition: n = 9/269</p>
Intervention group	<p>“Internet-based CBT” (with email) Setting: Internet-based and email Components: Interactive, self-guided Internet intervention to deliver family cognitive-behavioural therapy</p>
Comparison group	Waitlist control group
Outcomes	<p>Clinical significance: 50% or greater pre-post change on summed total pain score. Pain outcome: Clinically significant reduction (>50% reduction), mean pain intensity (NRS averaged over 0-10 days) Disability outcome: Child Activity and Limitations Interview (CALI) Depression outcome: Revised Child Anxiety and Depression Scale (RCADS). Anxiety outcome: Bath Adolescent Pain Questionnaire Treatment satisfaction: Treatment Evaluation Inventory</p>

Risk of bias table

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Low risk	Page 175 Randomization was implemented using a computer-generated randomization schedule to derive a randomization assignment to 2 treatment conditions in blocks of 4 for each ID number. The randomization assignment was programmed into the Web-MAP2 system
Allocation concealment (selection bias)	Low risk	Page 175 Randomization was implemented using a computer-generated randomization schedule to derive a randomization assignment to 2 treatment conditions in blocks of 4 for each ID number. The randomization assignment was programmed into the Web-MAP2 system into the Web-MAP2 system.
Blinding of participants and personnel (performance bias)	Low risk	Page 175 Participants were blinded to whether they were receiving an active or control treatment
Blinding of outcome assessment (detection bias)	Low risk	Measures completed and submitted online
Incomplete outcome data (attrition bias)	Low risk	PAGE 179 Attrition completely reported; non-significant differences between completers and non-completers were reported – “Participants who dropped out of the study and those who completed the study did not demonstrate statistically significant differences on any demographic variables or pre-treatment outcome variables (P values . 0.05)”.
Selective reporting (reporting bias)	Low risk	Data were fully reported
Other bias		

Rapoff 2014

Bibliographic information	Rapoff, M.A., Connelly, M., Bickel, J.L., Powers, S.W. Hershey, A.D., Allen, J.R., Karlson, C.W., Litzenburg, C. C. & Belmont, J.M. Headstrong intervention for pediatric migraine headache: a randomized clinical trial The Journal of Headache and Pain 2014, 15:12
Design of study (including randomisation procedure)	RCT. Two arms. Assessed at pre-treatment and post-treatment. Stratified randomisation. No description found in text.
Demographics	<p>Participants: N= 35 Sex: 25F, 10M Mean age = 10.2, 1.7 SD, range: 7-12 years Diagnosis = Headache Pain duration (months): not reported</p> <p>Recruitment: Headache clinic, 5.3 yr. recruitment period</p> <p>Analyses/Sample: ‘Intent to treat’ (LOCF): not reported</p> <p>‘As treated’: 35 Start of treatment: n = 35 Post-treatment: n = 22 Follow-up (three months): n = 22 Attrition: n = 13/35</p>
Intervention group	<p>“Computer-based CBT via CD-ROM” (and phone calls) Setting: Home computer Components: headache education, relaxation methods, problem-solving and stress management, targeted pain behaviour.</p>
Comparison group	Waitlist control group
Outcomes	<p>Clinical significance: 50% or greater pre-post change on summed total pain score. Pain outcome: Clinically significant reduction (>50% reduction), mean pain intensity (NRS averaged over 0-10 days) Disability outcome: Child Activity and Limitations Interview (CALI)</p>

	<p>Depression outcome: Revised Child Anxiety and Depression Scale (RCADS). Anxiety outcome: Bath Adolescent Pain Questionnaire Treatment satisfaction: Treatment Evaluation Inventory</p>
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Risk of bias table

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Unclear risk	Page 2 Participants were stratified by age (7–9 and 10–12) and randomly assigned following baseline to one of the two groups (education control or Headstrong).
Allocation concealment (selection bias)	Unclear risk	No description found in text
Blinding of participants and personnel (performance bias)	Unclear risk	No description found in text
Blinding of outcome assessment (detection bias)	Low risk	Measures completed at home and mailed back
Incomplete outcome data (attrition bias)	Unclear risk	Attrition completely reported; significant differences between completers and non-completers were not reported
Selective reporting (reporting bias)	Low risk	Data were fully reported
Other bias	Low risk	funded by a grant from the National Institutes of Health, (National Institute of Neurological Disorders and Stroke) R01-NS046641,

Stinson 2010

Bibliographic information	Stinson, J. N., McGrath, P. J., Hodnett, E. D., Feldman, B. M., Duffy, C. M., Huber, A. M., et al. (2010) An internet-based self-management program with telephone support for adolescents with arthritis: a pilot
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	randomized controlled trial. <i>Journal of Rheumatology</i> , 37(9), 1944–1952.
Design of study (including randomisation procedure)	Pilot RCT. Two arms. Assessed at pre-treatment and post-treatment. Fixed, block randomisation (blocks of 10) using an online random number generator. Concealed random allocation performed using an ID number in sealed envelopes.
Demographics	<p>Participants: N= 46 Sex: 31F, 15M Mean age = 14.7 (SD 1.6), range: 11-17 years Diagnosis = Juvenile idiopathic arthritis Pain duration (months): not reported</p> <p>Recruitment: recruited between October and November in 2008 from 4 pediatric tertiary care centres across Canada</p> <p>Analyses/Sample: ‘Intent to treat’ (LOCF): n= 46</p> <p>‘As treated’ = 43 Start of treatment: n = 43 Post-treatment: n = 37 Follow-up (three months): n = 37 Attrition: n = 6/46</p>
Intervention group	<p>“Internet-based CBT” (with email) Setting: Internet-based and email Components: education about arthritis, diagnosis, medications, managing symptoms, managing stress, negative thoughts, relaxation, distraction, self-care (exercise, nutrition, splints), self-monitoring, lifestyle issues, and looking ahead (education, vocation, transitional care issues).</p>
Comparison group	Attentional control group
Outcomes	<p>Pain outcome: Recalled Pain Inventory (RPI) Disability outcome: none Depression outcome: Revised Child Anxiety and Depression Scale (RCADS). Anxiety outcome: Bath Adolescent Pain Questionnaire Quality of life: Juvenile Arthritis Quality of Life Questionnaire (JAQQ). Treatment satisfaction: questionnaire developed by the investigators.</p>

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Trautmann 2010

Bibliographic information	Trautmann, E., & Kroner-Herwig, B. (2010) A randomized controlled trial of internet-based self-help training for recurrent headache in childhood and adolescence. <i>Behaviour Research and Therapy</i> , 48, 28–37.
Design of study (including randomisation procedure)	RCT. Three arms. Assessed at pre-treatment, post-treatment, six months. Randomisation using a computer-generated randomisation list. First author, using a randomly ordered list of groups to assign sequentially enrolled participants.
Demographics	Participants: N= 65 Sex: 36F, 30M Mean age = 12.6 (SD 2.1), range: 9-16 years Diagnosis = Headache Pain duration (months): NR Recruitment: Analyses/Sample: 'Intent to treat' (LOCF): NR 'As treated': 65 Start of treatment: n = 65 Post-treatment: n = 55 Follow-up (three months): n = 40 Attrition: n = 25/65
Intervention group	"Internet-based CBT" (with email) Setting: Internet-based and email Components: Interactive, self-guided Internet intervention to deliver cognitive-behavioural therapy: CBT: adapted from the manualized face-to-face group therapy program (Denecke and Kro"ner-Herwig, 2000) plus CD for use at home. AR: progressive relaxation, cue-controlled relaxation and differential relaxation. CD with

	these specific instruction tracks for the different stages of AR training to be used at home (4 tracks: a full relaxation protocol common to the CBT CD.
Comparison group	Education group received only the first self-help module (education on headache) + email contact. Participants in the wait-list control group continued with their standard care
Outcomes	<p>Clinical significance: 50% or greater pre-post change on summed total pain score.</p> <p>Pain outcome: Headache pain diary: VAS</p> <p>Disability outcome: none</p> <p>Depression outcome: Children's Depression Inventory,</p> <p>Anxiety outcome: Bath Adolescent Pain Questionnaire</p> <p>Quality of life: Health-related quality of life was assessed with the German KINDL-questionnaire (Ravens-Sieberer & Bullinger, 1998)</p> <p>Treatment satisfaction: developed by authors (0–3, higher scores signify a more positive evaluation for all three questions)</p>

Appendix 10: COREQ Checklist (Study 2)

Consolidated criteria for reporting qualitative research (COREQ) checklist for PRP groups

Item	Description
Domain 1: Research team and reflexivity	
Personal characteristics	
1. Interviewer/facilitator	AT and SOH
2. Credentials	AT: BA., MSc. SOH: PhD,
3. Occupation	AT: PhD candidate. SOH: Postdoctoral Fellow
4. Gender	AT: female. SOH: female
5. Experience and training	AT: trained in qualitative research methods and design; experience in conducting PRP groups; SOH: trained in qualitative research methods and design; has extensive experience in facilitating qualitative research groups including participative research process workshops.
Relationship with participants	
6. Relationship established	Potential participants contacted AT via email or telephone to discuss arrangements for the PRP groups. Otherwise participants had no relationship with researchers
7. Participant knowledge of the interviewer	Participants were informed that the researcher was conducting a PhD in the area of paediatric chronic pain management and that her goal was to understand paediatric pain self-management better by discussing it with people who lived with it. Participants were aware this research will be used to inform the content of an online intervention.
8. Interviewer characteristics	Qualitative researchers and supervisors were closely engaged in the research process and were therefore unable to completely avoid personal bias.
Domain 2: study design	
Theoretical framework	

9. Methodological orientation and Theory A participative research process approach and theoretical thematic analysis was used in this study.

Participant selection

10. Sampling Children who self-reported experiencing ‘chronic or recurrent pain lasting for a period of three months or more’ and their care-giver(s) were eligible to take part. Self-selected Irish JIA patients from the community participated in this research.

11. Method of approach From January to Jun 2015, chronic pain support groups and associations in the region were contacted. A press release was distributed to local media groups advertising the study (see Appendix 11).

12. Sample size There were 32 participants in the study. There were two child participative groups, five children took part in the first group and six children in the second. There were two parent groups, fifteen parents took part in the first group and six parents in the second.

13. Non-participation All participants who agreed on a date and time to attend took part in the groups.

Setting

14. Setting of data collection Data was collected in two separate locations, the first was a Family Fun Day event organised by Arthritis Ireland and the second was a meeting room in the School of Psychology at the University where the researcher is based.

15. Presence of non-participants No one else was present besides the participants and researchers.

16. Description of sample Demographic data can be seen in Table 4.2.

Data collection

17. Interview guide
Two of three open-ended questions were posed to each of the groups: (1) What things do you like to do when you are in pain? or (2) What things can you (your child) not do when you (your child) are in pain? and (3) “If it were your job to make life better for children with pain what would you do?”. These were the primary questions asked, with other probing questions offered during discussion: (1) What do you/we need to do that?, (2) “How do you/we do that? and (3) What support do you/we need to do that?
18. Repeat interviews
No repeat workshops were carried out.
19. Audio/visual recording
Audio recording was used to collect the data.
20. Field notes
Field notes were made during and after the PRP groups.
21. Duration
Each of the PRP groups was approximately 40 minutes in duration.
22. Data saturation
The researchers decided that data saturation had been achieved after the fourth PRP group. The written responses and audio transcripts were reviewed as soon as possible after each group session. Saturation was achieved as no further additional new information began to emerge. It was agreed that the addition of new categories by participants was unlikely after the fourth focus group (Fusch & Ness, 2015).
23. Transcripts returned
Written responses were not returned to participants for comment and/or correction. This was a multiphase process during which participants were afforded time to reflect on and/or revise their responses
- Domain 3: analysis and findings**
Data analysis
24. Number of data coders
Two data coders (AT and SOH) coded the data.
25. Description of the coding tree
Coding trees are presented in Chapter 4

26. Derivation of themes	Themes retrospectively coded based on an appropriate and fitting theoretical framework. The identified themes are reflective of the categories of responses identified by participants in the course of their data analysis. Two researchers agreed on a specified theoretical coding framework which guided the interpretation of the data (see Appendix 14.)
27. Software	Data were managed by hand
28. Participant checking	Participants did not provide feedback on the findings.
Reporting	
29. Quotations presented	Participant quotations were presented to illustrate the themes / findings. Each quotation identified using the participants' group and gender.
30. Data and findings consistent	There is consistency between the data presented and the findings. The unit of analyses was the theme rather than the prevalence or frequency of statements. Some statements of quantification are included (e.g. statements such as often, sometimes), but do not always aim to provide estimates of prevalence.
31. Clarity of major themes	In stage one, participants identified categories or themes which were reviewed by two study authors prior to theoretical thematic analysis. In stage two, these themes or categories were linked to pre-defined themes based on Social Cognitive Theory. A coding sheet was developed to guide this process. All major themes clearly presented in the findings.
32. Clarity of minor themes	There is a description of minor themes in the findings.

Appendix 11: Press Release for Qualitative Study (Study 2)

**NUI Galway Researchers Seek Participants For Project On
Chronic Pain In Children**

Children in Dublin, Cork, and Galway who experience chronic pain are invited to help develop an effective pain management programme

Researchers with the School of Psychology at NUI Galway are currently inviting children aged from 7-12 years who have experienced chronic or recurrent pain for a period of three months or more, and their parents, to help develop an effective pain management programme for young children which will be computer-based and accessed online.

Interviews will be run at several locations across Ireland including Dublin, Cork and Galway. Children with any type of chronic or persistent pain including abdominal pain, back pain, musculoskeletal pain, headache and combined or widespread pains, are invited to participate.

Participants will be shown a computer-based programme designed to support pain management and are invited to give feedback, based on their personal experiences of chronic pain management.

The project will involve either one meeting in groups of children and groups of parents or an individual interview in a convenient location, depending on preference. Your contribution will enable the researchers to decide what course of action would work best for this type of treatment programme. Group and individual interviews will be fun, informal and interactive, lasting 30-40 minutes.

Participant views and personal experiences are extremely valuable, and their input will greatly enhance research in the development of an intervention for children who suffer with chronic pain. They may also benefit from sharing their experiences and thoughts about chronic pain management with others in a similar situation, in a casual environment, while offering complete confidentiality.

NUI Galway researcher, Angeline Traynor said: “Chronic pain is increasingly prevalent in young people and can have a significant impact on the day-to-day quality of life. The most common types of chronic pain in children are abdominal pain, back pain, musculoskeletal pain, headache and combined or widespread pains.”

Ms Traynor continued, “Given the impact of chronic pain and the increasing focus on technology as a means of treatment delivery, it is essential to identify and address the needs of young children with respect to pain management. Participation is voluntary and anything you say during the group session is strictly confidential. These sessions will give children and their parents the chance to inform our research by telling us of their own personal experiences with chronic pain management.”

This programme is part of a PhD research project being carried out at NUI Galway by PhD student Angeline Traynor with Dr. Brian McGuire of the university’s School of Psychology and the Centre for Pain Research. The study is supported by Galway University Foundation.

If you would be willing to help with this important study or would like further information, please contact Angeline Traynor at a.traynor2@nuigalway.ie or 086 0378562 or go directly to www.helpkidswithpain.com

Appendix 12: Parent Participant Information Sheet and Consent Form
(Study 2)



LISTENING TO CHILDREN WITH PAIN

My name is Angeline Traynor, I am a Researcher with the School of Psychology at the National University of Ireland Galway. I would like to invite you to be part of a study about pain management for children with chronic pain and their parents or carers. We are asking you to work with us because your child experiences chronic or persistent pain.

Why do this study?

I would like to know about your experience of helping to manage your child's pain. Your views and personal experiences are extremely valuable. We believe you and your child are the experts in your own pain management and your opinion in your own words, will help us make a better pain management programme for children living with chronic pain.

What happens if I take part?

If you and your child agree to be part in this study we will be part of a group workshop where with parents and children will be asked about their everyday lives and experiences dealing with chronic pain, as well as their opinion about a pain management computer programme we are developing for children. This will involve just one meeting with a group of children and one meeting with a group of parents to share ideas and opinions about better pain management with us. You will be able to observe your child at all times. The researcher will ask questions like: *'what things do you do on a daily basis to help your child deal with their*

pain’ or *‘what do you think of the theme of our computer programme’?*

Group sessions will be fun and interactive and last no longer than 40 minutes. Refreshments will be provided.

Parents and children will be shown examples from a computer-based programme designed to support pain management and will be invited to give feedback, based on their personal experiences of chronic pain management.

To make the experience more enjoyable for your child they will be asked to draw some pictures or fill out some worksheets. As a thank you to the children for their time and to convey respect for their contribution we will all share some treats at the end.

Benefits?

We do not know if taking part will help you or your child. We think you will learn more about how psychological therapy can be used to cope with chronic pain and you will have a say in the design of a new computer-based pain management programme for children dealing with chronic pain. You may also benefit from sharing your experiences and thoughts about chronic pain management with others in a similar situation, in an environment which offers complete confidentiality. Your opinions will help us learn more about how the internet might be used to help children and parents deal with chronic pain.

Risks?

No serious risks of harm are expected however, it may be upsetting for you or your child to talk about how you cope with chronic pain on a daily basis and it may be upsetting to listen to how other parents or children discuss their experiences of dealing with chronic pain. If you feel you need to speak to someone about how you feel we would be happy to recommend someone and provide information on further sources of support. You do not have to answer any question that

you don't want to.

Confidentiality

Your identity will remain confidential. **Your name will be changed**, a pseudonym will be used to disguise your identity. The results may be published in a scientific journal so that others who support people with chronic pain can see what you have to say. Your name will not be published or disclosed to anyone outside the research team.

Who am I meeting?

You will be meeting researchers from the National University of Ireland Galway. The research team are specially trained and have been subject to security vetting by An Garda Síochána.

You Should Know

If you decide not to participate in this study your usual treatment will not be affected in any way. If you do decide to take part you are free to withdraw at any time without giving a reason.

The National University of Ireland Galway, Research Ethics Committee has reviewed and approved this study.

CONSENT FORM (STUDY 2)

- I confirm that I have received a copy of the Information Sheet for the above study. I have read it and I understand it. I have received an explanation of the nature and purpose and what my involvement will be.
- I have had time to consider whether to take part in this study and I have had the opportunity to ask questions.
- I understand that my participation is voluntary and that I am free to withdraw at any time, including after the information has been collected without giving

any reason.

- I understand that the information will be stored, on a confidential basis, on a computer and will be used for research purposes only.
- I understand that although I will have access to the information given by me, I will not have access to the information given by my child.
- I understand that my usual medical care will not be affected by our decision to participate or not.
- I understand the conversation that I and my child have with the researcher will be taped and that these tapes will be strictly confidential and used for analysis purposes only.

SIGNATURE OF PARENT OR LEGAL GUARDIAN

I agree to take part in the above study.

Yes No

I consent to my child taking part in the above study.

Yes No

You will be given a copy of this information to keep for your records.

Name of Child

(BLOCK CAPITALS PLEASE)

Name of Parent/Guardian

(BLOCK CAPITALS PLEASE)

Address of Parent/Guardian:

(BLOCK CAPITALS PLEASE)

Signature of Parent / Guardian:

Date:

Contact telephone:

SIGNATURE OF PERSON OBTAINING CONSENT AND PARENTAL PERMISSION

Name of Person Obtaining Consent and Permission

Signature of Person Obtaining Consent

Date:

Contact telephone:

Contact Information

If you would like any further information about the study, without any obligation to take part, please contact the researcher: Angeline Traynor, School of Psychology, National University of Ireland, Galway, Tel: +353-86 0378562 or email: a.traynor2@nuigalway.ie or visit: www.helpkidswithpain.com

Appendix 13: Child Participant Information Sheet and Assent Form
(Study 2)



HELP US HELP KIDS WITH PAIN!

My name is Angeline, I am called a researcher and I am part of a team who want to know what you think of a new computer programme we made to help children and their parents or carers deal with pain. We are asking you to work with us because you are between 7 and 12 years old and have pain that lasts a longtime.

What happens if I take part?

If you agree to take part in this study: You can choose to join a group of other children, the same age as you, who have pain like yours. There will be lots of art stuff and everyone will draw and talk about things they can do to make life better for children with pain. We will also show you a computer programme for children your age who have pain like yours. You and the other children can chat with the researcher about what you like and do not like about it.

Good Things

We hope you will learn more about how to deal with your pain and you can have a say about a new computer programme for children with pain like yours. With your help we will learn more about how the computer will help children and parents deal with pain. To thank you for being in the study, we will give you a small gift when you are finished.

Bad Things

We don't think that any big problems will happen if you join this study. You might feel sad when you talk about pain or when other children talk about their pain. If you want to talk to someone about your pain when you go home I can help you find someone.

Other things you should know

You do not have to answer any questions or do anything you do not want to. You do not have to take part if you don't want to. No one will be mad at you if you don't want to be in the study or if you join the study and change your mind later.

What do I do now?

Before you say **yes or no** talk to your Mum, Dad or whoever takes care of you about taking part. Ask someone to help you read this form. If you have any questions you can ask them any time.

- I know that my name will be changed and what I say and do will be private.
- I know that I can stop at any time.
- I know the talk I have with the researcher will be taped on a tape-recorder so she does not have to do a lot of writing

If you understand what is written, you can decide whether you would like to take part in the project.

I have decided that I would like to work with Angeline. Please put a circle around Yes or No.

Yes **No**

Please write your name below.

My name is:

My age is:

Thank you for taking the time to read about this study. You will be given a copy of this information sheet to keep.

Appendix 14: Social Cognitive Theory Coding Sheet (Study 2)

Bandura's (1986; 1997) Social Cognitive Theory Constructs and Definitions

Reciprocal determinism**Definition:**

"Social cognitive theory favours a model of causation involving triadic reciprocal determinism. In this model of reciprocal causation, behavior, cognition and other personal factors, and environmental influences all operate as interacting determinants that influence each other bidirectionally. Reciprocal causation does not mean that the different sources of influence are of equal strength. Some may be stronger than others. Nor do the reciprocal influences all occur simultaneously. It takes time for a causal factor to exert its influence and activate reciprocal influences." (Bandura, 1989, p. 2)

Psychological determinants**Self-efficacy****Definition:**

"Beliefs about personal ability to perform behaviors that bring desired outcomes" (Glanz, Lewis & Rimer, 2008, p.171). Self-efficacy is task-specific, meaning that self-efficacy can increase or decrease based on the specific task at hand, even in related areas. Among the types of thoughts that affect action, none is more central or pervasive than people's judgments of their capabilities to exercise control over events that affect their lives. The self-efficacy mechanism plays a central role in human agency (Bandura, 1982; 1986 as cited in Bandura, 1989, p. 59).

Outcome expectancies**Definition:**

Beliefs about the likelihood and value of the consequences of behavioral choices (Glanz, Lewis & Rimer, 2008, p.171). Outcome expectations refers to the physical, social, and self-evaluative expectations one holds for the outcome of one's behaviors. Outcome expectations also affect the level of self-efficacy. Individuals are more likely to engage in health behaviour activities if they believe the anticipated outcomes will be beneficial and that outcomes can be achieved. With increasing cognitive development, children become more skilled at judging probable outcomes of their actions. Such outcome expectations serve as incentives for observational learning (Bandura, 1989, p.29).

Behavioural determinants**Behavioural Goals****Definition:**

"In social cognitive theory, cognized goals, rooted in a value system, provide further self- incentives and guides to health behaviour. Goals may be distal ones that serve an orienting function, or proximal ones that regulate effort and guide action in the here and now. Intentions are essentially proximal goals. Both "I aim to do x" and "I intend to do x" refer to what a person proposes to do" (Bandura, 1986; Bandura 1998, p.7).

Self-regulation

Definition:

Controlling oneself through self-monitoring, goal-setting, feedback, self-reward, self-instruction, and enlistment of social support (Glanz, Lewis & Rimer, 2008, p.171). Success in self-regulation partly depends on the fidelity, consistency, and temporal proximity of self-monitoring. Self-observation serves at least two important functions in the process of self-regulation. It provides the information needed for setting realistic goals and for evaluating one's progress toward them (Bandura, 1986; Bandura 1998, p.12).

Incentive motivation

Definition:

The use and misuse of rewards and punishments to modify behavior (Glanz, Lewis & Rimer, 2008, p.171).

Environmental determinants

Observational learning

Definition:

"When viewed from the developmental perspective of social cognitive theory, observational learning is part of a more general process of cognitive and social development. But observational learning is also one of the basic means by which cognitive competencies are developed and expanded" (Bandura, 1989, p.30). "Learning to perform new behaviors by exposure to interpersonal or media displays of them, particularly through peer modeling" (Glanz, Lewis & Rimer, 2008, p.171). Observational learning is most effective when learners perceive that role models are like them (Glanz, Lewis & Rimer, 2008). Self-management learners are more likely to adopt new behaviors if their role models are similar to them in characteristics and managing a chronic disease (Sell, Amella, Mueller, Andrews, & Wachs, 2016).

Facilitation

Definition:

"Providing tools, resources, or environmental changes that make new behaviors easier to perform" (Glanz, Lewis & Rimer, 2008, p.171). "Social cognitive theory distinguishes between different types of barriers. Some of them are personal impediments that impede performance of the health behavior itself. They form an integral part of self-efficacy assessment" The regulation of behavior is not solely a personal matter. Some of the impediments to healthful living reside in health systems rather than in personal or situational impediments (Bandura, 1986; Bandura 1998, p.7-8).

Appendix 15: Parent Participant Information Sheet and Consent Form
(Study 3)



A SUPPORT PROGRAMME FOR CHILDREN WITH PAIN

My name is Angeline Traynor, I am a researcher with the School of Psychology at the National University of Ireland Galway. I would like to invite you to be part of a study about pain management for children with chronic pain. We are developing an online pain management programme for children with chronic pain and their care-givers and we would like your help.

Why do this study?

We are asking you to work with us because your child experiences chronic or persistent pain. Your views and personal experiences are extremely valuable. We believe you and your child are the experts in your own pain management and your opinion of our online programme, in your own words, will help us make a better pain management programme for children living with chronic pain.

What happens if I take part?

If you and your child agree to be part in this study, you and your child will be asked to use an online pain management programme on most days, for two weeks and then tell us what you think of it. You can share your ideas and opinions in one of two ways: (i) a group workshop where, in a separate group of parents and children, you will be asked your opinion about the programme or (ii) you can use the programme and complete an online questionnaire which asks what you think of it. The researcher will ask questions like: *‘what did you think of how the programme looked? How easy was the programme to understand?’*

Group sessions will be fun and interactive and last no longer than 30 minutes. Refreshments will be provided. To make the experience more enjoyable, you and your child will be asked to draw some pictures or use coloured paper to write your ideas. As a thank you to the children for their time and to convey respect for their contribution we will all share some treats at the end.

Benefits?

We do not know if taking part will help you or your child. We think you will learn more about how psychological therapy can be used to cope with chronic pain and you will have a say in the design of a new computer-based pain management programme for children dealing with chronic pain. You may also benefit from sharing your experiences and thoughts about chronic pain management with others in a similar situation, in an environment which offers complete confidentiality. Your opinions will help us learn more about how the internet might be used to help children and parents deal with chronic pain.

Risks?

No serious risks of harm are expected however, it may be upsetting for you or your child to talk about why you like or do not like parts of the programme based on how you cope with pain. If you feel you need to speak to someone about how you feel we would be happy to recommend someone and provide information on further sources of support. You do not have to answer any question that you don't want to.

Confidentiality

Your identity will remain confidential. **Your name will be changed**, a pseudonym will be used to disguise your identity. The results may be published in a scientific journal so that others who support people with chronic pain can see what you have to say. Your name will not be published or disclosed to anyone outside the research team.

Who am I meeting?

You will be meeting researchers from the National University of Ireland Galway. The research team are specially trained and have been subject to security vetting by An Garda Síochána.

You Should Know

If you decide not to participate in this study your usual treatment will not be affected in any way. If you do decide to take part you are free to withdraw at any time without giving a reason.

The National University of Ireland Galway, Research Ethics Committee has reviewed and approved this study.

CONSENT FORM (STUDY 3)

- I confirm that I have received a copy of the Information Sheet for the above study. I have read it and I understand it. I have received an explanation of the nature and purpose and what my involvement will be.
- I have had time to consider whether to take part in this study and I have had the opportunity to ask questions.

I understand that my participation is voluntary and that I am free to withdraw at any time, including after the information has been collected without giving any reason.

- I understand that the information will be stored, on a confidential basis, on a computer and will be used for research purposes only.
- I understand that although I will have access to the information given by me, I will not have access to the information given by my child.

- I understand that my usual medical care will not be affected by our decision to participate or not.
- I understand the conversation that I and my child have with the researcher will be taped and that these tapes will be strictly confidential and used for analysis purposes only.

SIGNATURE OF PARENT OR LEGAL GUARDIAN

I agree to take part in the above study.

Yes No

I consent to my child taking part in the above study.

Yes No

You will be given a copy of this information to keep for your records.

Name of Child

(BLOCK CAPITALS PLEASE)

Name of Parent/Guardian

(BLOCK CAPITALS PLEASE)

Address of Parent/Guardian:

(BLOCK CAPITALS PLEASE)

Signature of Parent / Guardian:

Date:

Contact telephone:

SIGNATURE OF PERSON OBTAINING CONSENT AND PARENTAL PERMISSION

Name of Person Obtaining Consent and Permission

Signature of Person Obtaining Consent

Date:

Contact telephone:

Contact Information

If you would like any further information about the study, without any obligation to take part, please contact the researcher: Angeline Traynor, School of Psychology, National University of Ireland, Galway, Tel: +353-86 0378562 or email: a.traynor2@nuigalway.ie or visit: www.helpkidswithpain.com

Appendix 16: Child Participant Information Sheet and Assent Form (Study 3)



A PROGRAMME FOR CHILDREN WITH PAIN

My name is Angeline, I am called a researcher and I am part of a team who want to know what you think of a new computer programme we made to help children and their parents or carers deal with pain. We are asking you to work with us because you are between 7 and 12 years old and have pain that lasts a long time.

What happens if I take part?

If you agree to take part in this study, we will show you a computer programme for children your age who have pain like yours. Next, we will ask you what you think of the programme. We will only ask questions about the programme, about what you think it needs to make it better. Questions like this: “what did you think of how the programme looked?”

If you would like to tell us what you think, you can do that in 2 ways:

1. you could join a group of children your age who have pain like yours and use art and coloured paper to draw or write your ideas

or

2. you could look at the programme and then use the computer in your home to answer questions and tell us what you think. The person who looks after you will be asked to help you do this.

Good Things

We hope you will learn more about how to deal with your pain and you can have a say about a new computer programme for children with pain like yours. With your help we will learn more about how the computer will help children and parents deal with pain.

Bad Things

We don't think that any big problems will happen if you join this study. You might feel sad when you think about how you deal with pain and why you liked or did not like different parts of the programme. If you want to talk to someone about your pain when you go home I can help you find someone.

Other things you should know

You do not have to answer any questions or do anything you do not want to. You do not have to take part if you don't want to. No one will be mad at you if you don't want to be in the study or if you join the study and change your mind later.

What do I do now?

Before you say **yes or no** talk to your Mum, Dad or whoever takes care of you about taking part. Ask someone to help you read this form. If you have any questions you can ask them any time.

- I know that my name will be changed and what I say and do will be private.
- I know that I can stop at any time.
- I know the talk I have with the researcher will be taped on a tape-recorder so she does not have to do a lot of writing

If you understand what is written, you can decide whether you would like to take part in the project.

I have decided that I would like to work with Angeline. Please put a circle around Yes or No..

Yes **No**

Please write your name below.

My name is: _____

My age is: _____

Thank you for taking the time to read about this study. You will be given a copy of this information sheet to keep.

Appendix 17: COREQ Checklist (Study 3)

Consolidated criteria for reporting qualitative research (COREQ) checklist for PRP groups

Item	Description
Domain 1: Research team and reflexivity	
Personal characteristics	
1. Interviewer/facilitator	AT, SOH
2. Credentials	AT: BA., MSc. SOH: PhD,
3. Occupation	AT: PhD candidate. SOH: Postdoctoral Fellow
4. Gender	AT: female. SOH: female
5. Experience and training	AT: trained in qualitative research methods and design; experience in conducting PRP groups; SOH: trained in qualitative research methods and design; has extensive experience in facilitating qualitative research groups including participative research process workshops.
Relationship with participants	
6. Relationship established	Participants contacted AT via email or telephone to discuss arrangements for the PRP groups and online survey. Otherwise participants had no relationship with researchers
7. Participant knowledge of the interviewer	Participants were informed that the researcher was conducting a PhD in the area of online paediatric chronic pain management and that her goal was to understand how such a programme might be used by and acceptable to children with chronic pain and their care givers. Participants were aware this research will be used to inform the content of an intervention.
8. Interviewer characteristics	Qualitative researchers and supervisors were closely engaged in the research process and were therefore unable to completely avoid personal bias.
Domain 2: study design	

Theoretical framework

9. Methodological orientation and Theory

PRP and theoretical thematic analysis was used in this study.

Participant selection

10. Sampling

Children who self-reported experiencing ‘chronic or recurrent pain lasting for a period of three months or more’ and their caregiver(s) were eligible to take part. Self-selected Irish JIA patients from the community participated in this research.

11. Method of approach

From May 2015 to Jan 2016, chronic pain support groups and associations in the region were contacted. Social media was the primary method of recruitment.

12. Sample size

There were 58 participants in the study. A total of 24 children and 34 parents contributed to the intervention website development process via a think aloud group, PRP workshop or online survey.

13. Non-participation

All participants who agreed on a date and time to attend took part in the think aloud and PRP groups.

Setting

14. Setting of data collection

Data was collected in separate locations using mixed methods. The first was a ‘think-aloud’ group session at a Family Fun Day event organised by Arthritis Ireland (Iteration 1); the second was a participatory research process workshop in the home of one participant (Iteration 2) and the third was an online survey following two weeks of access to the prototype programme (Iteration 3).

15. Presence of non-participants

No one else was present besides the participants and researchers.

16. Description of sample

Demographic data can be seen in Table 5.1 and 5.2.

Data collection

17. Interview guide In Iteration 1, the same questions were asked of children and parents: “What do you think of pain management support delivered using the Internet? / “What do you think of this method of delivery (Feeling Better programme examples)”. In Iteration 2, the same questions were asked of children and parents: “If you (your child) were to use this website / take part in this programme - what would it need to have to keep your interest / help you (your child) to cope with pain?”. In Iteration 3, a series of qualitative open-ended questions based on Ritterband’s behaviour change model for internet interventions (Ritterband et al., 2009) was posed. Participants were asked to complete an online survey assessment of their user experience. The online survey included demographic characteristics and the Internet Evaluation and Utility Questionnaire (Thorndike et al., 2008; Ritterband et. al., 2008).
18. Repeat interviews No repeat workshops were carried out.
19. Audio/visual recording Audio recording was used to collect the data.
20. Field notes Field notes were made during and after each session.
21. Duration Each of the PRP groups was approximately 30 minutes in duration.
22. Data saturation The researchers decided that data saturation had been achieved following the online survey completed in Iteration 3. Saturation was achieved as no further additional new information began to emerge.
23. Transcripts returned Written responses were not returned to participants for comment and/or correction. This was a multiphase process during which participants were afforded time to reflect on and/or revise their responses

Domain 3: analysis and findings

Data analysis

24. Number of data coders Two data coders (AT, SOH) coded the data.

25. Description of the coding tree	In place of a coding tree, this data is linked to theoretical concepts, cognitive behavioural strategies and mechanisms of behaviour change in a matrix used to guide intervention development
26. Derivation of themes	Themes were retrospectively coded based on an appropriate theoretical framework. The identified themes are reflective of the categories of responses identified by participants in the course of their data analysis.
27. Software	Data were managed by hand
28. Participant checking	Participants did not provide feedback on the findings.
Reporting	
29. Quotations presented	Participant quotations were presented to illustrate the themes / findings. Each quotation identified using the participants' group and.
30. Data and findings consistent	There is consistency between the data presented and the findings. The unit of analyses was participant identified themes rather than the prevalence or frequency of statements. Some statements of quantification are included (e.g. statements such as often, sometimes), but do not always aim to provide estimates of prevalence.
31. Clarity of major themes	Participants identified codes or categories of responses. These categories were treated as codes with which to refine theoretical analysis. All major themes clearly presented in the findings.
32. Clarity of minor themes	There is a description of minor themes in the findings.

Appendix 18: Parent Participant Information Sheet and Consent Form
(Study 4)

***FEELING BETTER: AN INTERNET-BASED PAIN
MANAGEMENT PROGRAMME FOR CHILDREN WITH
CHRONIC PAIN.***

Thank you for your interest in the *Feeling Better* research programme. Before you decide whether you would like to be part of this research project, it is important that you understand why we are carrying out this research and what it will involve.

What is this study all about?

The aim of the study is **to expand the knowledge and range of chronic pain coping skills used by school age children with chronic pain**. To do this we have developed an internet- based pain-management programme called *Feeling Better* which we would like your help to evaluate. This programme aims to (i) help school age children with chronic pain to gradually increase their level of activity and manage their pain and (ii) to help you as their care-giver to guide your child through the process of pain self-management. We want to test the programme by comparing two groups of children with pain: one group who receive their usual medical care plus the *Feeling Better* intervention (Internet Group A) and another group who receive medical care as usual only (Control Group B). A research project of this nature has not been investigated to date in Ireland.

Who can take part in this study?

You are invited to take part in this study if all the following are true:

- You have a child aged 5-12 years who has experienced chronic or persistent pain for a period of three months or more.
- You have regular access to a computer and to the internet;

- You are willing to abstain from any new psychological treatment for chronic pain during the active phase (9 weeks) of this study;
- Your child's chronic pain condition is not due to malignancy (e.g. cancer)
- You and your child have adequate English language ability.

What does this study involve?

If you and your child agree to take part, you will be asked to complete a consent form and questionnaire. The questionnaire will feature a **parent and child section** which **each take about 10 minutes to complete**. After you have returned the questionnaire by clicking SUBMIT on the website page, you will automatically receive a message telling you if you are suitable to take part in the study. Your suitability will be based on your child's age and particular level of disability. If you are suitable to take part in the research, you will be assigned at random to one of two conditions: the internet intervention condition (Group A) or the waitlist control condition (Group B). This means the research team will not choose which part of the study your child is in. Though participation is voluntary, children who complete the programme will be rewarded with a gift voucher for their contribution be entered into a draw to win €100.00 as a reward for their contribution.

Internet Group A

If you and your child are assigned to 'Internet Group A', your child will take part in the *Feeling Better* training programme. As the care-giver, you will be guided through an information-based, complementary section of the programme. For children, each weekly session will involve information about chronic pain and instruction on a range of cognitive and behavioural techniques. For care-givers, each weekly session will involve information about chronic pain and instruction on how to guide your child toward pain self-management. The programme has **9 sessions** which were designed by clinical psychologist who is trained in pain

management for children. Each weekly session will focus on a different topic and range of techniques. Behaviour therapy sessions will involve relaxation training and activity pacing to improve physical function. Cognitive therapy sessions will help identify negative thinking patterns and improve coping skills. You will be asked to complete **one session each week. Each session will take approximately 30 minutes to complete.** The programme will be delivered using a secure website protected by industry standard encryption. You will be asked to complete a questionnaire featuring a parent and child section at the beginning and again at the end of this 9-week programme. This questionnaire will take about 10 minutes to complete each section. **No aspect of your usual care will be affected**

Control Group B

If you and your child are assigned to ‘Control Group B’ you will **not initially have access** to the *Feeling Better* programme. You will be asked to complete a questionnaire at the beginning and again at the end of a 9-11 week time period. These questionnaires will take about 10 minutes to complete. This **comparison information** will help us decide if the intervention is acceptable for children with pain. **No aspect of your usual care will be affected.** When these questionnaires have been completed, after a period of approximately 9 weeks, you and your child will then be invited to take part in the *Feeling Better* programme. Thus, everybody who registers for this research project will have the opportunity to benefit from the online pain management programme.

At approximately four months after completing the initial questionnaire, participants in both groups will be asked to complete a final brief questionnaire.

Benefits?

We think you will benefit from access to a **free online source of support** and information relating to chronic pain management. Further

benefits may follow from training in cognitive behaviour therapy techniques **specifically tailored** for chronic pain management and for school age children. You will be helping us to test the usability of a programme designed to help children and their care-givers cope with chronic pain. When this research project is concluded, all participants who have completed the programme will receive a summary of the main findings. It could take in excess of 12 months before final results are published.

Risks?

As part of this study you and your child will be asked to complete a questionnaire at 3 different time points. This will include questions that ask about your feelings/moods and what your child thinks about pain. For some children or parents, reflecting on these questions may be upsetting. However, we do not anticipate these questions will cause significant distress. These questionnaires have been used in many studies previously. It is not expected that any potential risks should arise regarding your or your child's participation. In the unlikely event that completing the questionnaire makes you or your child feel uncomfortable, you are free to withdraw at any time from the research or you can also choose not to answer those specific questions. You and/or your child might find while you are answering them that you would like to talk to someone about some of the issues raised. We will be happy to recommend someone.

If I Say No

Your participation is entirely voluntary. If you and your child decide not to participate in this study your treatment will not be affected in any way. If you and your child decide to take part you are still free to withdraw at any time and without giving a reason. Your rights will not be affected in any way by your decision.

Confidentiality

Your identity will remain confidential. All children and families participating in this study will be given a participant number. This number will be used to identify all of the information we get so your name will not be on any data forms. Only your consent form will have your name and this information will be stored separately from all the other materials. The only circumstances where the researchers cannot guarantee confidentiality is if it is discovered during the course of the research that a child is at risk of harm.

Has this study received Ethics Committee Approval?

This study is organized by the National University of Ireland Galway. The National University of Ireland Galway, Research Ethics Committee have reviewed and approved this trial.

Where can I get further information?

A report detailing the research findings will be available from the researcher following the research's completion. You and your child may choose to be included on a mailing list to receive a copy of this report.

Contact Information.

If you would like any further information about the study, without any obligation to take part, please contact the research co-ordinator: Angeline Traynor, School of Psychology, National University of Ireland, Galway, Tel: 086 037 8562 or email: team@feelingbetter.ie

If you have any queries relating to the research, the researchers can be contacted at the following e-mail addresses: Angeline Traynor – a.traynor2@nuigalway.ie or team@feelingbetter.ie

National University of Ireland, Galway:

Dr Brian McGuire 091- 493454 or brian.mcguire@nuigalway.ie

If you have any concerns about the research and wish to speak to someone in confidence, you can contact the Head of School, Dr. Annmarie Groarke at 091-493101.

Thank you for taking the time to read this information sheet.

CONSENT FORM (ONLINE)

Please select the ‘Yes’ Option only if all the following are true:

I confirm that I have read the Participant Information for this project.

- Yes
- No

I have read the information sheet and I understand the purpose, duration, foreseeable risks and benefits and what my involvement will be.

- Yes
- No

I understand that my participation is voluntary and that I am free to withdraw at any time, without my medical care being affected.

- Yes
- No

I agree to take part in the *Feeling Better* study. Please choose only one

of the following:

- Yes
- No

I consent to my child taking part in the *Feeling Better* study. Please choose only one of the following:

- Yes
- No

I consent to be contacted following this study for the purpose of follow-up research. Please choose only one of the following:

- Yes
- No

Appendix 19: Child Participant Information Sheet and Assent Form
(Online) (Study 4)

***FEELING BETTER: A PROGRAMME FOR CHILDREN WITH
PAIN THAT LASTS A LONG TIME.***

What and Why?

A research study is a way to learn more about something. In this study **we want to see if the Internet programme we made for children who have pain like yours is helpful and if children like using it.**

You Should Know.

You get to decide if you want to be in this study. **You can say ‘No’ or you can say ‘Yes’ and whatever you decide is OK.** We’d like you to know that:

- If you say ‘Yes’ - you can always change your mind later.** 
- If you say ‘No’ - no one will be upset with you.** 
- Your usual care will carry on as normal, whatever you decide.**
- You can stop at any time.** 

There are some reasons why we would ask that you stop being in the study: (1) if you do not try to learn or practice or (2) if we feel that stopping the study is important for your safety.

What would happen if I join the study?

If you decide to be in the study: **we would need to ask you a few questions, like:**

- Are you feeling happy or sad?
- What things do you find easy to do?
- What things do you find hard to do?

This is how we find out if the study is a right for you. We will then tell you if it is still OK for you to be in the study. You would need to answer these questions at three different times in the study, at the beginning and again at the end. It should take about 10 minutes to answer these questions. You will be put in **'Internet Group A'** or **'Control Group B'**.



If you are in **Internet Group A**, you would use *Feeling Better*, an Internet programme that teaches about pain and about different things you can do to help you deal with pain. This programme has 9 different parts that show you different skills to deal with about pain. You would be asked to do one part each week for 9 weeks. Each part takes about 30 minutes to do.

If you are in **Control Group B**, you would be part of what is called a control group. You will be asked to answer questions at three different times in the study before you will be allowed to join. This means you would have to wait awhile before you will be able to join the Internet programme. If you are in **Group B**, we will tell you when it is your turn to take part in the internet programme.

About three months after your stop the programme you will be asked to answer a few more questions about how you feel and what things you find hard or easy to do.

Good Things?

We think being in this study will **help you by teaching you skills** to deal with pain and how it makes you feel. You can feel good about helping us to test this programme and make it better for children your age who have pain like yours.

Bad Things?

We don't think that any big problems will happen to you if you join this study, but **you might feel sad when you are answering questions** about how you are feeling. If you want to talk to someone we will help you find someone to talk to about how you feel.

Private

Your name will be changed to a number and no one outside the study team will know it. The **answers you give will be kept secret**. We will not tell your doctor or your family what you say. Everything used in the study will be kept safe and private. But, if you tell me something that is very serious, like somebody is in trouble or somebody has harmed you, we may have to tell someone.

To thank you for being in the study, we would give you a small gift when you finish the programme.

If I have questions who do I ask?

You can ask the researcher, Angeline, any questions you have at any time. If you have a question later that you didn't think of now, you can **send her an email message: team@feelingbetter.ie**

Remember:

- Your doctors will treat you whatever you decide**
- You can ask your family or friends for help** on this.
- You can stop at any time** and you won't get into trouble.

- You can say 'No' or you can say 'Yes'
- Whatever you decide will be OK.
- If you say 'Yes', you can always change your mind later.

CONSENT FORM (ONLINE: Post Parent Consent)

Your parents (carers) knows about this study and that we are asking if you would like to be part of it.

If you **do** want to take part in this study you can pick the **YES** button.

If you **do not** want to be part of the study you can pick the **No** button

Please pick the 'Yes' button only if all the next sentences are true:

I know that the answers I give will be kept private and that my name will be changed to a number.

- Yes
- No

I know that I can stop the study at any time. Please choose only one of the following:

- Yes
- No

I want to be in the study. Please choose only one of the following:

- Yes
- No

Appendix 20: Press release for Pilot Randomised Controlled Trial
(Study 4)

**NUI Galway Researchers Seek Participants for Online Pain
Management Programme**

*Children who experience chronic pain are invited to take part in an
online pain management programme at NUI Galway*

Monday, 18 January, 2016: Researchers from the School of Psychology and Centre for Pain Research at NUI Galway are currently recruiting children age 6-10 years with chronic pain and their parent(s) or care-giver(s) to take part in an online pain management programme for children.

An online pain management programme called *Feeling Better* has been developed at NUI Galway to help children and parents to manage chronic pain for a better quality of life. This web-based programme is based on the principles of cognitive behavioural therapy, a psychological therapy which has shown to be effective in the management of chronic pain, in traditional face-to-face therapy and group treatment. The *Feeling Better* study is unique in that a trial of this nature has not been investigated to date in Ireland. The programme is currently the only, widely available, source of interactive, online therapeutic support for school age children with chronic pain in Ireland. The researchers would like to enlist families coping with chronic pain to aid in the testing of this online pain management programme.

Chronic pain is pain which persists for a period of three months or more. It affects up to 35% of the Irish population and is increasingly prevalent in young people. Recent studies suggest up to 10% of 5-12 year old Irish children report chronic or persistent pain including abdominal pain, back pain, musculoskeletal pain, headache and

widespread pain. Chronic pain is often associated with psychological effects, which may include changes in mood, difficulty focusing attention and performance at school. This can have a significant impact on day-to-day quality of life.

The *Feeling Better* study is open to children with any type of chronic or persistent pain (pain which has lasted for three months or more). The study will take place over the coming months and children and their parent(s) from across Ireland and internationally are invited to take part. Pain support groups, parent-led networks, GPs and physiotherapists around the country are encouraged to get in touch and to refer suitable people with pain to the study. Benefits to participants include access to a free online pain management programme and training in cognitive and behavioural techniques tailored for chronic pain management and school age children.

The online programme was developed by clinical psychologists and researchers at NUI Galway with input from families currently coping with chronic pain. School age children with chronic pain and their care-givers were involved in the design and development process. Evidence-based psychological strategies were selected to address areas of pain management children and parents identified as most challenging and important. This influence ensures *Feeling Better* is a fun and engaging form of online therapeutic support designed by children with pain for children with pain.

The programme involves 9-weekly online sessions. Each session is designed to take approximately 30-minutes to complete and all participants are guided through the programme by a ‘Coach’ who is available to provide feedback and advice on a regular basis. Each week, this fun, pirate-themed, interactive programme will introduce children to new skills in the form of ‘Challenges’ and weekly ‘Missions (treatment sessions)’ which they must complete in order to progress in their training.

Participating children will begin the programme as a ‘Powder Monkey’ and must earn a promotion with each Mission until they succeed to ‘Captain’ and claim their treasure (small reward). Parents are encouraged to take the role of ‘Coach’ and are separately guided through a complementary section of the programme where they are provided with information, tips for practice and tools to help with day-to-day pain management.

Weekly sessions are tailored to participants goals, support needs and coping preferences. Children and parents will learn more about psychological strategies which focus on such as relaxation training, activity pacing, attention management, communication skills and the influence of thoughts and emotions on the experience of pain.

This programme is part of a research project being carried out at NUI Galway by PhD candidate and Hardiman scholar, Angeline Traynor and led by Professor Brian McGuire from NUI Galway’s School of Psychology and the Centre for Pain Research. Angeline Traynor has been researching chronic pain management and working with families to develop an effective and accessible pain management programme.

Ms Traynor says: “Chronic pain is thought to be predictive of long term complaints and disability. Given the impact of chronic pain it is essential to provide a means of support for young children with respect to pain management. Learning coping strategies at an early age may have long term benefits for the child and the family as a whole. Our hope is that this online programme will overcome access and resource issues which may be preventing families from receiving psychological treatment to support pain management.”

Participation is voluntary. Children and parents who take part will be helping researchers decide if web-based technology is an acceptable means of treatment delivery. The researchers are looking for

volunteers to help them trial the programme and determine what works and what doesn't work.

To participate in the study or for further information, please contact Angeline Traynor at team@feelingbetter.ie and 086 0378562 or visit www.feelingbetter.ie

The study is supported by Galway University Foundation and the Centre for Pain Research at NUI Galway.

ENDS

Contact Gwen O'Sullivan, Acting Press and Information Executive, NUI Galway on 091 495695 or gwen.osullivan@nuigalway.ie

Appendix 21: Recruitment Poster (Study 4)

WANTED! CHILDREN WITH CHRONIC PAIN AND THEIR PARENTS

Try out our free, online Feeling Better programme to help children cope with pain: www.feelingbetter.ie

What?
This study is about using the internet to learn new ways to cope better with pain

Who?
Children, 5-12 years, who have pain that has lasted for longer than 3 months / Parents who have a child with chronic pain

How?
Go online for 30 minutes a week for 9 weeks

Interested?
Go to: www.feelingbetter.ie
Or Contact us for more information:
team@feelingbetter.ie
Tel: 086 037 8562

This study has been approved by the National University of Ireland Galway, Research Ethics Committee

Galway University FOUNDATION

PAIN Research

Pain In Child Health
Strategic Training Initiative in Health Research
Children's Institute of Health Research

NUI Galway OÉ Gaillimh

Appendix 22: Certificate of Completion (Study 4)



Appendix 23: Questionnaires for intervention (Study 4)

Questionnaire – Parents**Demographic Characteristics – Baseline assessment only**

1. Please enter your first name:
2. Please enter your last name:
3. Please enter your email address:
4. What is your relationship to this child? Please choose only one of the following:
 - Mother, Step Mother, Foster Mother
 - Father, Step Father, Foster Father
 - Grandmother
 - Grandfather
 - Guardian
 - Other
5. Is your child currently undergoing any other form of psychological therapy for chronic pain? Please choose only one of the following:
 - Yes
 - No
6. Is your child currently experiencing a psychiatric illness? Please choose only one of the following:
 - Yes
 - No
7. Does your child have cognitive deficits, a learning disability or any other issue which would prevent him or her from being able to read and understand the research questionnaire? Please choose only one of the following:
 - Yes
 - No
8. Is your child's chronic pain due to malignancy (e.g. cancer-related illness)? Please choose only one of the following:
 - Yes

- No
9. Throughout our lives, most of us have had pain from time to time (such as minor headaches, sprains, and toothaches). Does your child have pain other than these everyday kinds of pain most days? Please choose only one of the following:
- Yes
 - No
10. How long has your child suffered from chronic pain? Please choose only one of the following:
- Less than 3 months
 - Less than 6 months
 - Less than 1 year
 - 1-2 years
 - 2-5 years
 - More than 5 years
11. Do you have regular access to a personal computer and the internet? Please choose only one of the following:
- Yes
 - No

Information about your child – Baseline assessment only

12. Please enter your child's first name:
13. Please enter your child's last name:
14. Please enter your child's age:
15. Child is - Please choose only one of the following:
- Female
 - Male
16. Ethnic Group or Race - Please choose only one of the following:
- White Irish
 - White Irish Traveller
 - Any other White background
 - Black or Black Irish - African
 - Black or Black Irish - Any other Black background
 - Asian or Asian Irish - Chinese
 - Asian or Asian Irish - Any other Asian background

17. What is the name of your child's chronic pain condition (if known)? Please write your answer here:

Information about you - (Baseline assessment only)

18. Gender - Please select your gender:
- Female
 - Male
19. Marital Status - Please choose only one of the following:
- Single
 - Married
 - Separated
 - Living with someone
 - Divorced
 - Widowed
20. What is the highest degree or level of schooling you have completed? Please choose only one of the following:
- Some primary or secondary school
 - Finished secondary school
 - Some post-secondary education
 - Third level degree
 - Master's degree or above
21. Employment Status - Please choose only one of the following:
- Paid employment - full time
 - Paid employment - part time
 - Voluntary work - unpaid
 - Sheltered work
 - Registered as unemployed but available for work
 - Retired
 - Student
 - Housewife/husband
 - Other
22. Country of residence * Please enter your country of residence:
23. If Ireland, which county do you live in?
24. Treatment Expectation - How much do you think this programme might help your child? NRS-11: 0 is Not Hard at all

and 10 is Very, Very Hard

25. Pain Intensity - (Baseline, post-treatment and follow-up)



Each face shows a person who has no pain (hurt), or some, or a lot of pain. Face 0 doesn't hurt at all. Face 2 hurts just a little bit. Face 4 hurts a little bit more. Face 6 hurts even more. Face 8 hurts a whole lot. Face 10 hurts as much as you can imagine, although you don't have to be crying to have this much pain. Please choose the face that shows the pain you think your child is feeling.

- Please choose the face that shows your child's pain at its worst in the last 2 weeks.
- Please choose the face that shows your child's pain at its least in the last 2 weeks.
- Please choose the face that shows how much pain your child has on average (as it usually is, most days)
- Please choose the face that shows how much pain your child has right now

26. How often does your child feel pain? Please choose the appropriate response for each item:

- 1-2 times per week
- 3-6 times per week
- Daily

27. About how many days in the past two weeks did your child have no pain (pain free)? Please choose the appropriate response for each item:

0	1	2	3	4	5	6	7
8	9	10	11	12	13	14	

28. Think about the days that your child had pain in the last 2 weeks. Please choose the appropriate response for each item:

- Less than 15 minutes –
- Less than 30 minutes –
- Less than 1 hour –
- Between 1-2 hours –
- All morning –
- All afternoon –
- All day

29. On average, how long has your child's usual level of pain lasted?

30. On average, how long has your child's worst level of pain lasted?

31. On average, how difficult is it for your child to fall asleep or stay asleep at night because of pain?

Standardised Measures Used – Parent Form - (Baseline, post-treatment and follow-up)

32. Pain Intensity was measured using the Wong-Baker FACES Pain Rating Scale (Wong-Baker FACES Foundation, 2019)

33. Health-related quality of life was assessed using the Pediatric

Quality of Life Inventory – Parent report (PedsQL™ 4.0; Varni, 1998).

34. Use of strategies to cope with pain was measured using the Pediatric Quality of Life Inventory – Coping Skills Inventory – Parent report (PedsQL-CSI; Varni, 1996).

35. Level of catastrophizing was assessed with the Pain Catastrophising Scale- Parent report (PCS-C&P; Crombez et al., 2003).

36. Pain related self-efficacy was assessed using the Self-efficacy for Functioning Despite Pain Scale –Parent report (Bursch et al., 2006).

37. Levels of parental protective behaviour was assessed using the Adult Response to Children’s Symptoms–Protect Subscale- Parent report (Walker, Ley & Whitehead, 2006).

Child Questionnaire

Getting to Know You – (Baseline assessment only)

1. What is your name? Text box
2. What age are you? Text box
3. Are you a boy (male) or a girl (female)? Please choose only one of the following:
 - girl • boy
4. What class or year are you in at school? Please write your answer here:
5. Do you know the name of your pain problem? Please write your answer here:
6. About how long have you had a pain problem (felt pain or hurt)? Please choose only one of the following:
 - Less than 3 months
 - Less than 6 months
 - Less than 1 year
 - 1-2 years
 - 2-5 years
 - More than 5 years
 - Other
7. Pain Location - Where do you feel pain or hurt? You can choose more than one answer.
 - Head
 - Arms
 - Hands
 - Chest
 - Tummy / Belly
 - Back
 - Neck
 - Shoulders
 - Hips
 - Legs

- Feet
- Knees
- Ankles
- Other:

8. Click the number that shows how much you think this programme might help you? 0 is No Help and 10 is the Most Help you can imagine:

0 1 2 3 4 5 6 7 8 9
10

9. Pain or Hurt – (Baseline, post-treatment and follow-up assessment)



Each face shows a person who has no pain (hurt), or some, or a lot of pain. Face 0 doesn't hurt at all. Face 2 hurts just a little bit. Face 4 hurts a little bit more. Face 6 hurts even more. Face 8 hurts a whole lot. Face 10 hurts as much as you can imagine, although you don't have to be crying to have this much pain. Please choose the face that shows the pain you are feeling.

- Please choose the face that shows your pain or hurt at its highest (worst) in the last 14 days (2 weeks)
- Please choose the face that shows your pain or hurt at its lowest (least) in the last 14 days (2 weeks)
- Please choose the face that shows your pain or hurt as it usually is (most days)

- Please choose the face that shows how much pain or hurt you have right now

10. How often do you feel pain? Please choose the appropriate response for each item:

- 1-2 times per week
- 3-6 times per week
- Daily

11. How often do you feel pain? In the past 2 weeks (14 days) how many days did you have no pain (pain free)? Please choose the appropriate response for each item:

0 1 2 3 4 5 6 7 8
9 10 11 12 13 14

12. On a normal day, how long does your (usual) pain or hurt last?

13. On the days when you have the most (worst) pain or hurt, how long does it usually last?

- 1-2 hours
- 3-5 hours
- 5 or more hours
- morning
- afternoon
- evening
- all day

14. Sleep - Click on the number that shows how hard it is for you to fall asleep or stay asleep at night because of pain? NRS-11: 0 is Not Hard at all and 10 is Very, Very Hard

Standardised Measures Used – Age Appropriate (5-7 years and 8-12 years)

Child Form:

15. Pain Intensity was measured using the Wong-Baker FACES Pain Rating Scale (Wong-Baker FACES Foundation, 2019)

16. Health-related quality of life was assessed using the Pediatric Quality of Life Inventory – Child report Child report (5-7 and 8-12 years) (PedsQL™ 4.0; Varni, 1998).

17. Use of strategies to cope with pain was measured using the Pediatric Quality of Life Inventory – Coping Skills Inventory – Child report (5-7 and 8-12 years) (PedsQL-CSI; Varni, 1996).

18. Level of catastrophizing was assessed with the Pain Catastrophising Scale - Child report (PCS-C&P; Crombez et al., 2003).

19. Pain related self-efficacy was assessed using the Self-efficacy for Functioning Despite Pain Scale – Child report (Bursch et al., 2006).

20. Levels of parental protective behaviour was assessed using the Adult Response to Children’s Symptoms–Protect Subscale- Child report (Walker, Ley & Whitehead, 2006).

21. Internet Evaluation and Utility Questionnaire – Post-treatment only

These questions are about your use of the web programme, *Feeling Better*. Please read the items and tell us how you felt about using the *Feeling Better* programme. If the item does not apply, please choose “NA”.

1. How easy was the *Feeling Better* programme to use?

<i>Not at all</i>	<i>Slightly</i>	<i>Somewhat</i>	<i>Mostly</i>	<i>Very</i>	<i>NA</i>
0	1	2	3	4	5

2. How convenient was the *Feeling Better* programme to use?

<i>Not at all</i>	<i>Slightly</i>	<i>Somewhat</i>	<i>Mostly</i>	<i>Very</i>	<i>NA</i>
0	1	2	3	4	5

3. How much did the *Feeling Better* programme keep your interest and attention?

<i>Not at all</i>	<i>Slightly</i>	<i>Somewhat</i>	<i>Mostly</i>	<i>Very</i>	<i>NA</i>
0	1	2	3	4	5

4. How much did you like the *Feeling Better* programme?

<i>Not at all</i>	<i>Slightly</i>	<i>Somewhat</i>	<i>Mostly</i>	<i>Very</i>	<i>NA</i>
0	1	2	3	4	5

5. How much did you like the way the *Feeling Better* programme looked?

<i>Not at all</i>	<i>Slightly</i>	<i>Somewhat</i>	<i>Mostly</i>	<i>Very</i>	<i>NA</i>
0	1	2	3	4	5

6. How worried were you about your privacy in using this *Feeling Better* programme?

<i>Not at all</i>	<i>Slightly</i>	<i>Somewhat</i>	<i>Mostly</i>	<i>Very</i>	<i>NA</i>
<i>0</i>	<i>1</i>	<i>2</i>	<i>3</i>	<i>4</i>	<i>5</i>

7. How satisfied were you with the Feeling Better programme?

<i>Not at all</i>	<i>Slightly</i>	<i>Somewhat</i>	<i>Mostly</i>	<i>Very</i>	<i>NA</i>
<i>0</i>	<i>1</i>	<i>2</i>	<i>3</i>	<i>4</i>	<i>5</i>

8. How good of a fit was the Feeling Better programme for you?

<i>Not at all</i>	<i>Slightly</i>	<i>Somewhat</i>	<i>Mostly</i>	<i>Very</i>	<i>NA</i>
<i>0</i>	<i>1</i>	<i>2</i>	<i>3</i>	<i>4</i>	<i>5</i>

9. How useful did you find the information in the Feeling Better programme?

<i>Not at all</i>	<i>Slightly</i>	<i>Somewhat</i>	<i>Mostly</i>	<i>Very</i>	<i>NA</i>
<i>0</i>	<i>1</i>	<i>2</i>	<i>3</i>	<i>4</i>	<i>5</i>

10. How easy was the information to understand?

<i>Not at all</i>	<i>Slightly</i>	<i>Somewhat</i>	<i>Mostly</i>	<i>Very</i>	<i>NA</i>
<i>0</i>	<i>1</i>	<i>2</i>	<i>3</i>	<i>4</i>	<i>5</i>

11. How much did you feel you could trust the information?

<i>Not at all</i>	<i>Slightly</i>	<i>Somewhat</i>	<i>Mostly</i>	<i>Very</i>	<i>NA</i>
<i>0</i>	<i>1</i>	<i>2</i>	<i>3</i>	<i>4</i>	<i>5</i>

12. If difficulties continue or return, how likely would you be to come back to this Feeling Better programme?

<i>Not at all</i>	<i>Slightly</i>	<i>Somewhat</i>	<i>Mostly</i>	<i>Very</i>	<i>NA</i>
<i>0</i>	<i>1</i>	<i>2</i>	<i>3</i>	<i>4</i>	<i>5</i>

13. *How good of a method was the Internet for delivering this intervention?*

<i>Not at all</i>	<i>Slightly</i>	<i>Somewhat</i>	<i>Mostly</i>	<i>Very</i>	<i>NA</i>
<i>0</i>	<i>1</i>	<i>2</i>	<i>3</i>	<i>4</i>	<i>5</i>

14. *What was the most helpful part of the Feeling Better programme?*

15. *What was the least helpful part of the Feeling Better programme?*