### Title
Quality of Life Issues: Juvenile Idiopathic Arthritis and Young People in Ireland

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Quality of Life Issues: Juvenile Idiopathic Arthritis and Young People in Ireland

Submitted in part fulfillment of the academic requirements for the degree of Doctor of Philosophy (PhD)

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Submitted to: Discipline of Health Promotion
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Table of Contents

Abstract .................................................................................................................................................. 1
Declaration ............................................................................................................................................... 2
Acknowledgements ............................................................................................................................... 3

1. INTRODUCTION ................................................................................................................................. 4
1.1 Background ......................................................................................................................................... 6
1.2 Aims and Objectives of the Study ....................................................................................................... 7
1.2.1 Objectives: .................................................................................................................................... 7
1.3 Rationale for the Study ....................................................................................................................... 8
1.4 Summary of Chapters ........................................................................................................................ 9

2. LITERATURE REVIEW .......................................................................................................................... 13
2.1 Introduction ...................................................................................................................................... 13
2.1.1 Literature Search .......................................................................................................................... 13
2.2 The Concept of Quality of Life ........................................................................................................ 14
2.3 The Medical Approach/Health-Related Quality of Life Approach ..................................................... 16
2.3.1 Health-Related Quality of Life (HRQOL) in Young People ............................................................ 23
2.3.2 Health-Related Quality of Life – Measurement Issues ................................................................. 26
2.4 Disease-Specific Questionnaires for Juvenile Arthritis ..................................................................... 30
2.4.1 The Childhood Health Assessment Questionnaire (CHAQ) ......................................................... 30
2.4.2 Juvenile Arthritis Functional Assessment Scale and Report ........................................................ 31
2.4.3 Juvenile Arthritis Self-Report Index .............................................................................................. 31
2.4.4 Juvenile Arthritis Quality of Life Questionnaire ............................................................................. 31
2.4.5 Childhood Arthritis Health Profile ............................................................................................... 32
2.4.6 Paediatric Rheumatology International Trials Organization Clinical Assessment .................................. 32
2.4.8 The DISABKIDS Health-Related Quality of Life Measure ............................................................ 33
2.5 Summary ......................................................................................................................................... 36
2.6 What is Juvenile Idiopathic Arthritis? ............................................................................................... 38
2.6.1 Types of JIA ................................................................................................................................. 42
2.6.2 Oligoarticular JIA ....................................................................................................................... 42
2.6.3 Polyarticular JIA .......................................................................................................................... 44
2.6.3.1 Polyarthritis (rheumatoid factor negative) ............................................................................... 44
2.6.3.2 Polyarthritis (rheumatoid factor positive) ............................................................................... 45
2.6.4 Systemic Arthritis ...................................................................................................................... 45
2.6.5 Psoriatic Arthritis ........................................................................................................................ 46
2.6.6 Enthesitis-Related Arthritis ...................................................................................................... 46
2.7 Principles of Management of the Disease .......................................................................................... 48
2.8 Pain Assessment and Management .................................................................................................. 51
2.9 Psychosocial Impact of Juvenile Idiopathic Arthritis ....................................................................... 56
2.10 Healthcare Strategies for Young People with Chronic Diseases ..................................................... 65
2.11 Communications ............................................................................................................................. 66
2.12 Social Support ................................................................................................................................ 71
2.13 Transition to Adult Services .......................................................................................................... 75
Section One

Introduction

The Organization of Care

The Impact of JIA on Social Support on Quality of Life

Quality of Life: Social Support for the Family

Quality of Life: Parental Involvement

Quality of Life: Psychological Well-being

Quality of Life: Physical Well-being

Quality of Life: the Global Well-being

Quality of Life: Referral to a Regional Rheumatology Centre

Quality of Life: the Impact on the Family

Quality of Life: the Impact of the Diagnosis Process

Quality of Life: the Impact on the Young Person

Quality of Life: The Impact of Illness on Family Life

Quality of Life: Social Acceptance, Normalcy, and Moving On

Quality of Life: the Impact of the Diagnosis Process, Physically and Psychologically

Quality of Life: Parents as Advocates for their Children

Quality of Life: Referral to a Regional Rheumatology Centre

Quality of Life: the Global Well-being of the Young Person

Quality of Life: Diagnosis and the Impact on the Family

Quality of Life: The Impact of Illness on Family Life

Quality of Life: Medication

Quality of Life: Family Support

Results of Interviews with Parents

Results of Interviews with Parents

Diagnosis

Young People

Medication/Treatment

Communication

Young Adults and Transition from Juvenile Services to Adult Services

The Healthcare Service for Young People with Juvenile Idiopathic Arthritis

The Role of the Team

Summary

DISCUSSION

Introduction

The Impact of JIA on the Bio-psychosocial Well-being and Quality of Life on a Young Person with Juvenile Idiopathic Arthritis (JIA)

The Impact of JIA on Family Quality of Life

The Impact of Social Support on Quality of Life

The Organization of Care
Appendices

APPENDIX 1: Tannahill’s Model of Health Promotion
APPENDIX 2: Differential Diagnosis of Joint Pain In Children
APPENDIX 3: Potential Complications of Juvenile Idiopathic Arthritis
APPENDIX 4: Cytokine Modulators Currently In Use or Under Assessment for the Treatment of Severe JIA
APPENDIX 5: Garda Clearance
APPENDIX 6: Ethical Approval from Hospitals
APPENDIX 7: Interview Schedule with Young People
APPENDIX 8: Information for Participants with Arthritis (a)
APPENDIX 9: Interview Schedule for Parents
APPENDIX 10: Topics for Discussion with Clinicians
APPENDIX 11: Consultant Survey
APPENDIX 12: Data from Three Perspectives
APPENDIX 13: An Example of Physical and Personal
APPENDIX 14: An Example of Coping Resources/Skills Needed By a Young Person with JIA
APPENDIX 15: Analogy of the Pit Stop Crew
Tables

Table 2.1: Characteristics of JIA ................................................................. 43
Table 4.1: Phases of Thematic Analysis .................................................. 116
Table 5.1: Health and Activity ............................................................... 124
Table 5.2: Psychological Well-being ..................................................... 125
Table 5.3: Moods and Emotions ............................................................. 127
Table 5.4: Leisure Time ........................................................................ 128
Table 5.5: Time with Parents ................................................................. 129
Table 5.6: Finance ............................................................................. 129
Table 5.7: Friends ............................................................................. 131
Table 5.8: School ............................................................................. 132
Table 5.9: The Impact of Arthritis .......................................................... 133
Table 5.10: Quality of Life: Medical Treatment ...................................... 135
Table 5.11: Questionnaire: Quality of Life About Symptoms of the Disease .............................................................................. 136
Table 5.12: Questionnaire: Part E About Your Healthcare Services ....... 138

Figures

Figure 1.1: An Overview of the Research Study Process.......................... 12
Figure 4.1: Phase One ....................................................................... 95
Figure 4.2: Phase Two ....................................................................... 97
Figure 4.3: Phase Three ..................................................................... 102
Figure 4.4: Adapted from the DISABKIDs Group (2001) 1 ...................... 106
Figure 4.5: Adapted from the DISABKIDs Group (2001) 2 ...................... 107
Figure 4.6: Phase Four ..................................................................... 108
Figure 4.7: Phase Five ...................................................................... 114
Figure 5.1: Key Issues to Emerge from the Study .................................. 186
Figure 5.2: Demonstrating Convergence and Non-Convergence of Findings .................................................................................... 187
Figure A: An Example of a ‘Micro-context’ Model of Care for Young People with JIA ........................................................................................................ 348
Abstract

The aim of this study was to explore quality of life issues of young people in Ireland with juvenile idiopathic arthritis (JIA). JIA is an autoimmune inflammatory disease diagnosed during childhood before the age of 16 with an incidence of 10-20/100,000. It is characterized by persistent synovial inflammation which can cause functional impairment, pain, and activity limitation; all of which may affect quality of life. Treatment modalities are not curative and aim to control the inflammatory process. This was a mixed methods study. To obtain multiple perspectives, young people with JIA were surveyed and parents and clinicians were interviewed.

Findings

The young people perceived that they had a good quality of life. Social support was found to be a protective factor in buffering the impact of juvenile arthritis. Parents had a strong role in buffering their children from adverse circumstances and promoting their children’s quality of life.

Parental recognition of the practical difficulties that children were encountering and pride in their efforts to adapt to difficult situations was a major theme. Education, school services and future prospects were frequent issues. Socialization was considered very important. However, services and service organization for young people with JIA were frequently encountered as problematic. Safety considerations relating to the long-term effects of the newer biologic medications, and whether they had made the right choices for their children, were significant concerns for parents.

Conclusions

Person-centred care from an early age is needed to meet the needs of young people with JIA across their lifespan, to optimize their life opportunities and quality of life. Service provision and service organization needs to be more collaborative and co-ordinated. A dedicated liaison person is needed to coordinate and support young people with JIA and their families to manage and adapt to the illness and improve their quality of life. Finally, families need more knowledge and information relating to the newer biologic medications.
Declaration

National University of Ireland, Galway

Title: Quality of Life Issues: Juvenile Idiopathic Arthritis and Young People in Ireland

Name: Mary O’Hara

Student Identity Number: 98976893

Date Submitted:

I hereby declare this work is entirely my own and that I have acknowledged the writings, ideas and work of others.

Furthermore I have not knowingly allowed another to copy my work.

Signature: ___________________________ Date: __________
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1. INTRODUCTION

Juvenile idiopathic arthritis (JIA) is a complex, chronic, inflammatory group of conditions that begin in childhood and involve persistent inflammation in one or more joints. JIA can cause serious complications in physical function and can significantly affect the well-being of the young person. Classification of JIA continues to evolve, resulting in better knowledge and understanding of patient subgroups. The incidence of the disease is approximately 1 in 10,000 and the prevalence is 1 in 1000 (Friswell 2004; Ravelli & Martini 2007; Beresford 2011). Pain, stiffness, physical function limitations and psychosocial issues may impact on the health-related quality of life of the young person. Impaired joint function, joint destruction, reduced growth and eye problems are potential complications. The goal of treatment for JIA is to suppress the disease, control it, achieve remission, prevent damage to joints, and promote the normal parameters of growth and well-being. Chronic pain and loss of education can impact on career choices, life prospects and employment opportunities (Shaw, et al., 2006).

Reducing the psychosocial impact of this condition is of critical importance to strengthen social inclusion and avoid marginalisation and isolation. Normal daily activities of life can be strained, constrained and constricted. Young people who had previously been independent may require assistance with basic activities of living such as dressing, eating and bathing. As a consequence of disease fluctuation, symptoms can vary not only from day to day but also throughout the day. Symptoms can be subtle, often, children and young people do not complain of symptoms, highlighting the need for thorough clinical assessment (Altschuler, 1997). Disease management is often complex (Wallace, 2010). The condition frequently necessitates visits to specialized medical services. These can impact on the young person’s social time, leisure activities and education (Sallfors et al., 2002). Many of these young people have been attending hospital outpatient clinics and inpatient units from an early age and frequently have a well-developed personal knowledge of the healthcare system.
Concerns about altered body image, social acceptance, independence and future prospects may have an impact on the health-related quality of life of young people with JIA (Barlow et al., 1999). Other commentators suggest that educational attainment is generally equivalent to that of peers; however, marriage and employment are less (Packham, 2002; Arkela-Kautiainen, 2005).

Two health promotion models were used to provide a framework for this study and support it: Tannahill’s model (Downie et al., 1996) and The Innovative Chronic Care Framework (WHO, 2003). Tannahill’s model (Downie et al., 1996) illustrated by three overlapping spheres of activity: health education, health protection and prevention of ill health (please see Appendix 1). Health education provides information and opportunities to influence attitudes and well-being. Health protection is concerned with safeguarding health. The concept also allows for the prevention of co-morbidities. This may be done through a number of approaches including fiscal, social and legislative. Prevention of ill-health is concerned with reducing or avoiding the risk of ill health through the use of medical strategies and environmental or social interventions (Naidoo & Wills, 2000). The concept of health protection was incorporated into this model, which allowed for the development during adolescence of the young people living with this chronic disease. This concept captured the vulnerability of the group being studied; for example, the majority were under the age of 18 and legally were considered as minors. The model also had the capacity to appreciate the dynamics of the trajectory of the disease, and through health education it could encapsulate the need for knowledge and information, preventing ill health and enhancing and promoting health and well-being. This model also allowed for a pluralistic approach to this exploratory study. The Innovative Chronic Care Framework (WHO, 2003) provided a three-level approach to the study using: a micro-level, a meso-level and a macro-level approach health promotion model to further support the study.
A combination of qualitative and quantitative research methods was used in the study. It was applied research using a pluralistic approach and the study was underpinned by a pragmatist paradigm.

1.1 Background

The concept of health promotion is a broad one. It aims to assist people as individuals or groups to optimize their health and well-being by enabling them to take more control of their own health through knowledge and education. It is viewed not as an event but as a process, which promotes healthy behaviours and endeavours to prevent ill-health (WHO, 1984). It can also be considered as a set of values or principles (Naidoo & Wills, 1998). Promoting health is impacted by many factors including socio-economic, environmental and political, and is strongly influenced by the co-ordination, co-operation and interagency partnerships of many disciplines.

Health promotion is also related to the prevention of illness. In general terms, the health of the world is improving, life expectancy is increasing, infectious diseases are declining and infant mortality rates continue to decrease. People are living longer due to many factors, including improved nutrition and hygiene. Advances in technology, and developments in medicine and healthcare provision, have also had a role in improved life expectancy (WHO, 2006). However, in recent years there has been a marked increase in the prevalence of chronic diseases (WHO, 2007). The impact of these conditions can significantly affect the quality of life of the person with the condition, and also others within the social circle (Cadman et al., 1991, Thorpe et al., 2009). These conditions require complex responses from patients and healthcare providers over an extended period of time, often decades (Unwin et al., 2004). Frequently, the conditions are labile or there are intercurrent acute episodes that can add to the complexity of the situation (Nolte & McKee, 2008). Where a cure is not possible, maximising quality of life is an important health promotion goal for this population.
In the context of adolescence, a time of immense and unique change biopsychosocially, a young person with a chronic health condition is confronted with many challenges that can impact on their quality of life. Challenging issues that can impact on the young person include: the provision of treatment for and comprehensive management of their chronic disease, the visibility of the disease, social issues, education, lifestyle, mobility and communication issues (WHO, 2007). The chronic condition affecting adolescents to be considered in this work is the auto-immune disease of juvenile idiopathic arthritis (JIA).

1.2 Aims and Objectives of the Study
The aim of the study was to explore the health-related quality of life of young people in Ireland with juvenile idiopathic arthritis (JIA).

1.2.1 Objectives:

➢ To explore the health-related quality of life of young people with JIA and compare their views with a peer group.
➢ To explore the health services experiences of young people with JIA.
➢ To explore the perceptions of quality of life issues of parents of young people with juvenile idiopathic arthritis.
➢ To explore the perspectives of clinicians in respect of quality of life issues for young people with JIA.
1.3  Rationale for the Study

The rationale for exploring the quality of life of a group of young people in Ireland with the specific chronic disease of juvenile idiopathic arthritis was that, as a group, they were under-investigated. Currently this chronic disease is not curable but it can be suppressed. An informal request to access information relating to the condition and services in Ireland launched a search for information. A registry of young people in Ireland with the disease was not available at that time; consequently it was not known how many young people in the country had this condition. The characteristics of this particular disease include a tendency to lack visibility; often it is insidious but it is not benign (Duffy 2006). These facts were particularly interesting to me. I wanted to know how young people coped with this condition and how it impacted on their lives and their quality of life. Treatment protocols and effective health services can also impact on global well-being and quality of life I was interested if these were issues for young people with arthritis and if so, were their issues being addressed. A paucity of information and literature relating to young people with the condition and available services in Ireland provided a further impetus to develop a deeper interest in the subject area, as little was known in Ireland about the specific needs and quality of life of individuals with JIA.

Relevant Terms

To avoid repetition the words adolescent, teenager, child, children, young person and young people have been used interchangeably, and all refer to individuals aged approximately 12 to 18 years.
1.4 Summary of Chapters

Introduction

This section introduces the research study, its aims and objectives, and the rationale for the study.

Chapter 1 – Literature Review

Chapter one reviews the literature and explores the definition and origins of the terms ‘quality of life’ and ‘health-related quality of life’. The concept was found to be complex and multidimensional, needing a holistic perspective to understand it in context as it can change over time. Definitions were categorized as global, component, focused and combination definitions. The chapter outlines the available health-related quality of life research evaluation instruments for application with young people with juvenile idiopathic arthritis. Many of these instruments were developed to focus on physical function only, while others measure quality of life in addition to physical function. Some of the instruments are very detailed, providing excellent data; however, they could take an unduly long time to complete, which may cause some difficulty with administration and response rate. Generic measures may provide information on global well-being but may not be sufficiently sensitive to a particular population. The second part of the chapter discusses the heterogeneous disease of juvenile idiopathic arthritis, its signs and symptoms, its management and its biopsychosocial impact. The importance of communication and social support are reviewed, the process of transition from paediatric services to adult rheumatology services is examined.
Chapter 2 - Methodology

This chapter considers the ethical issues relating to the project. The research was concerned with a vulnerable population of young people under the age of eighteen. Issues relating to child protection, consent and assent are examined. Best research practice issues relating to this population are discussed. Data were collected from adults too; therefore ethical issues relating to adults are also examined. Gaining access to research sites and the requirements of their research ethics committees are also considered. The second part of the chapter discusses the philosophical underpinnings of the study, including the health promotion models used to structure and support the study. The exploratory nature of the research question shapes the theoretical framework, methods and sampling of this quality of life study. The design of the study needed to be fit for purpose, and this is also discussed. Issues that need to be addressed include the appropriateness of the methodology to answer the research question, and whether the methodology would add value to the study or weaken it. Triangulating the data and the methods allow for the potential of complementary data to enhance the understanding of the phenomena under study. The ‘real-world’ context and external issues to the study further promoted a pragmatic approach to the theoretical framework of the study and are considered in this chapter.

Chapter 3 - Methods

This chapter details the methods used to collect the data relating to the quality of life of young people with JIA. Two research methods are used in the design of this study. Quantitative survey instruments are used with consultant paediatricians, consultant rheumatologists, adolescents with JIA, and their peer group without arthritis. Qualitative interviews are used with the young people to generate questions to use in a health services questionnaire, and also with parents and specialist clinicians. The chapter highlighted and discusses the operational issues and challenges relating to the data collection.
Chapter 4 - Findings

In this chapter the findings of the study are reported. Key findings from the five phases of the study are:

**Phase 1:** Seventy percent of clinicians responded that they did not have any patients with juvenile idiopathic arthritis.

**Phase 2:** Themes to emerge from the interviews with the young people included issues relating to communications with healthcare providers, proficiency of healthcare providers, access to allied healthcare services, free medications, and co-ordination of services.

**Phase 3:** The young people perceived that their quality of life was good, also it compared favourably with a comparator group.

**Phase 4:** Key quality of life issues to emerge from interviews with parents included service organization, treatment management, social support, pride in their children, and concern related to the long-term impact of medications.

**Phase 5:** The main issues to emerge from the clinician interviews related to treatment management, good communications with the young person and the family, education and adapting to the illness, and the need for more co-ordinated service provision. They acknowledged that as the young people grow into adults, new issues emerge and the quality of life issues change.

Chapter 5 - Discussion

The discussion addresses quality of life issues for the young people in the study with JIA. Adapting to and coping with the illness are discussed. The importance of maintaining social interaction with peers and having fun with friends while having this unpredictable chronic disease of JIA were emphasized. Social support and social connection were considered strong components of quality of life and mediating factors in coping with the disease. The discussion addresses the significance of a health promotion model to structure the study. The study design combined qualitative and quantitative approaches. These broadened the scope of the study and gained overlapping perspectives from respondents. The importance of education at all levels and across the lifespan was considered for the young people, parents and clinicians. Service organization, communication
and the concept of partnership in patient-centred care were explored. Parents spoke of the impact of the situation on the quality of family life. They spoke of how service organization could be improved, but also of the positive work of clinicians. Transition to adult services, which was a concern of parents and clinicians, is examined. Looking forward, a model of care for young people with JIA is proposed.
Figure 1.1: An Overview of the Research Study Process
2. LITERATURE REVIEW

2.1 Introduction

The purpose of this chapter is to review the literature relating to quality of life and juvenile idiopathic arthritis. The aim is to provide an understanding of the concept of quality of life and of the disease of juvenile idiopathic arthritis.

The first three sections of this chapter are specific to quality of life and discuss the concept of quality of life, health-related quality life approaches and disease-specific quality of life questionnaires for juvenile idiopathic arthritis. These sections are followed by sections relating to the disease of juvenile idiopathic arthritis and its bio-psychosocial impact on a person with the condition, issues relating to the principles of management of the disease including pain assessment and management; pharmacological interventions; social support and; transition to adult services.

2.1.1 Literature Search

Literature searches were undertaken of the following electronic databases: Online EBSCO host, PubMed (1966-December 2011), CINAHL (1982-December 2011), EMBASE (1980-December 2011). The following terms were used in the search strategy: quality of life, health-related quality of life issues, juvenile idiopathic arthritis (JIA), juvenile rheumatoid arthritis (JRA), juvenile chronic arthritis (JCA), juvenile arthritis (JA), uveitis, chronic disease, chronic illness. As the study evolved other areas of the literature were searched using words and phrases such as: adaptation to chronic illness, stigma, communicating with adolescents, social support and discursive othering.

The language was restricted to English. Books, journals and newspapers were hand searched also.
2.2 The Concept of Quality of Life

There are many definitions of the term ‘quality of life’ and these are reflected in the approaches to the study of the concept. Currently there is no universally agreed definition. Often it is used in terms of objective and subjective indicators and is associated with other concepts such as happiness, well-being and functional status. This chapter aims to provide a background to the concept, and to consider the main theoretical approaches to quality of life and the evolving definitions of the concept.

Historically, philosophical references were made to ‘quality’ in ancient Greek and Chinese literature when questions were posed such as, ‘how is quality affected by conflicts and challenges?’ In more recent times, the advent and adoption of new technologies, new specialist areas, new products and changing demographics have had a profound effect on the way people live and work. The economic approach to quality of life is concerned with economic growth. This is measured in terms of gross national product. Using this approach measurable indicators and outcomes can be objectively assessed and evaluated with a view to improving systems. On an individual basis this is measured in terms of acquired material wealth and its impact on the many dimensions of life. However, the economic approach recognizes that affluence without happiness does not constitute a good quality of life and what constitutes happiness is not easily measured (Eiser & Morse, 2001a).

When utilizing an economic approach a quantitative situation does not necessarily include relevant qualitative issues that are important to the individual to indicate their perception and interpretation of a good quality of life. It has been recognized that objective external indicators need to be enhanced by subjective assessment, which would provide more qualitative data, potentially providing a more comprehensive assessment of quality of life and an individual’s circumstances.
Maslow’s (1954, 1962) hierarchy of human needs framework provides a theoretical model to assist in evaluating quality of life and considers variables such as hunger, thirst, loneliness and security. Other variables such as learning, mastery and self-actualization are also considered by Maslow as significant areas of human need and have implications for the assessment of quality of life (Higgs et al., 2003). Many of these variables are expressed in both sociological and psychological approaches to quality of life.

The expression ‘quality of life’ has its origins in a sociological approach to determine the standard of life of people in the United States. Expanding on this construct other integral aspects of life have been considered to be essential to quality of life. These include life satisfaction, self-esteem, well-being, general health, functional status, and life adjustment (Lovell & White 1990). The sociological approach is concerned with the social and environmental issues of quality of life. It emphasizes the significance of relationships and culture (Eiser & Morse, 2001a). Bradford and Larson (2002) suggest the advantage of this approach is that there is potential to highlight developmental and environmental aspects of life that may influence quality of life.

The sociological approach is strongly connected to the psychological approach to quality of life, which stresses the importance of high self-esteem, autonomy, control, coping, self-sufficiency, personal happiness fulfilment and social competence (Abbey & Andrews, 1986; Fry, 2000). Other models, known as classic models, assess subjective well-being, happiness, morale and life satisfaction (Andrews & Withey, 1976; Larsen, 1978; Andrews, 1986). Social expectations or gap models are based on the disparities between the individual’s real life situation and their preferred or aspirational circumstances (Calman, 1984; Michalos et al., 2001). The ‘lived experience’ of the individual and their unique appreciation of their life situation support a phenomenological model of quality of life (O’Boyle, 1977). This model is supported by Ziller (1974), Benner (1985) and Rosenberg (1995) who add to the construct by recognising not only the importance of the individual’s unique experience of life.
and its quality but also how the value system of the individual will influence their quality of life.

2.3 The Medical Approach/Health-Related Quality of Life Approach

The medical approach or health-related quality of life approach emanated from advances in medical science. New technologies, medicines and treatment modalities have enabled people to survive life-threatening diseases, chronic diseases and traumatic injuries and crises that would have been fatal in previous times. It was considered important to measure how people were surviving in terms of functional ability and to determine, following therapeutic treatment/intervention, if they felt better (Moons et al., 2006). Other reasons for measuring outcomes included comparing outcomes in clinical trials, commissioning programmes of care and attaining the point of view of the individual to assess patient satisfaction with service delivery. A range of approaches can be utilized, including elements from the economic, psychological and sociological approaches. Health-related quality of life can be measured at both an individual level or group level and can be both objective or subjective to assess many domains, including physical functioning, psychological status and social status.

Health-related quality of life approaches have been used in many adult areas of ill health. Initial interest was related to the functional impact of a disease (Abeles et al., 1994; Binner 1991). Other work extended into the impact of medical treatments and rehabilitation (Spilker, 1990; Hollandsworth, 1988). Increasingly, in addition to functional ability, quality of life approaches are gaining insights into the impact of the illness and its treatment on physical, mental and social well-being (McDowell & Newell, 1987; Stewart & Ware, 1992; Bowling, 1991, 2005; Bowling et al., 2003).

The debate relating to the significance of objective assessment tools and their sensitivity versus subjective assessment tools continues in health-related quality of life. Day and Jankey (1996) assert that assessing objective
perspectives of quality of life is important, but these are outweighed by subjective perspectives and evaluation of quality of life. Social support is recognized as an important dimension of health and well-being. Cobb (1976:300) defines social support as ‘information leading the subject to believe that he or she is cared for and loved, is esteemed and valued, and belongs to a network of communication and mutual obligation’. Health is thought to be affected by social support through positive psychological and behavioural processes (Uchino, 2006). It is thought that social support can act as a buffer, which assists the individual to cope or adapt to stressful life events.

Initially health-related quality of life placed emphasis on physical and functional ability until the late 1990s/2000s. Quality-adjusted life years (QALYs), that is life expectancy for quality of remaining years, could also assist in decision making and allocation of resources. These measures tend to be illness-based and to be utilized after the event. The medical approach is strongly influenced by all of the approaches previously discussed and now endeavours to comprehensively evaluate the impact of an illness on an individual (Eiser & Morse, 2001a).

Thus a psychological and sociological perspective focusing on the wider determinants of health integrated into a health-related approach to quality of life can provide a broader framework in which to examine quality of life (Raphael, 2000). Bowling (2005), acknowledging the depth and breadth of the concept, suggests that quality of life is about the ‘goodness of life’ in all its levels, and health is one aspect impacting on quality of life.

The World Health Organization (WHO) defined the term health as ‘a state of complete physical, mental and social well-being, and not merely the absence of disease or infirmity’ (WHO 1948). Health is influenced by many factors including level of education, income, environment and access to health services. The WHO definition of health has strongly influenced the defining of quality of life by many authors. Ware (1984) and Aaronson (1988) added other domains
including disease state, physical symptoms, functional status, psychological functioning and social functioning, expanding further the definition of quality of life. Spieth and Harris (1996) observe that the physical, mental and social dimensions noted in the WHO definition continue to remain core to the concept of quality of life. However, the WHO definition was revised in 1998 to reflect the greater complexity of the concept. Bowling (2005) suggests that a broad model is needed to encapsulate the many dimensions of quality of life and health-related quality of life.

The World Health Organization Quality of Life Group state ‘quality of life is defined as individuals’ perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns’. It takes into consideration ‘physical health, psychological state, level of independence, social relationships, personal beliefs and their relationships to salient features of the environment’ (The WHOQOL Group, 1995:15).

Raphael (1998:1) suggests that in simple terms quality of life means ‘How good is life for you?’ He makes the point that each person has unique complex and multidimensional life experiences, and consequently each person has an individualistic approach to quality of life, making it difficult to define. Measuring quality of life, in Raphael’s estimation, needs to be holistic and individualistic. It does, however, need to include functional ability such as to care for oneself, and mobility, as these are strongly correlated with self-efficacy and independent living in the community. Self-efficacy is recognized increasingly as a vital component in positive wellbeing. For example, a person with arthritis may limit their physical activities through fear of pain. This can lead to regression of activity and in a ‘domino response’ this can impact on their physical abilities by reducing their physical conditioning and leading to the potential for other co-morbidities.
Raphael’s (1998) Quality of Life Profile explores three domains of life. These are ‘Being’, ‘Belonging’ and ‘Becoming’. These concepts consider the importance of social relations, social integration and social support and their impact on health. ‘Being’ - who you are as a person - includes the physical, psychological and spiritual ‘Being’. ‘Belonging’ – how you fit in with people and places includes physical, social and community belonging. ‘Becoming’ – things you do in your life that define you includes practical, leisure and potential growth opportunities. It also includes adjustments that the person may need to make to their lifestyle and their aspirations for the future, who they are to ‘Become’ in the future.

Carr et al., (2001:1240) acknowledge that the definition of quality of life is a dynamic one and that in relation to the new significant issues of health in the 21st century the definition of quality of life is likely to be revised again. ‘The way we think about health and health care is changing. The two factors driving this change are the recognition of the importance of the social consequences of disease, and the acknowledgement that medical interventions aim to increase the length and quality of survival. For these reasons, the quality, effectiveness and efficiency of health care are often evaluated by their impact on a patient’s ‘quality of life’.’

Other commentators broadened their perspective, for example Marinelli and Plummer (1999) included the physical, social, emotional, intellectual, spiritual and environmental in their perspective on quality of life.

A quality of life perspective can help to inform health policy. Consequently, quality of life as a concept has become significantly more important in health promotion and rehabilitation. The principles of health promotion include participation, empowerment, holism, equity, multi-strategy, intersectoral and sustainability (WHO 1998). To promote health, appropriate public policies, health initiatives and interventions need to be in place. In order to enact policies and provide these services knowledge and insights about particular
conditions and their impact need to be generated. Zissi et al., (1998) suggest that quality of life can be influenced by multiple co-related factors such as health, well-being and functional status. They are of the view that quality of life can be ‘mediated’ by other ‘inter-related’ factors such as self-efficacy and self-esteem. Windle et al., (2011:2), defining resilience, suggest that ‘it is the process of negotiating, managing and adapting to significant sources of stress or trauma’. Assets and resources within the individual, their life and environment facilitate this capacity for adaptation and ‘bouncing back’ in the face of adversity. They suggest that resilience can vary across the life span. Gilligan (2000) suggests that young people facing adversity can be enabled to be more resilient if their self-esteem and self-efficacy are supported by others demonstrating their caring and valuing of them.

Browne, et al., (1997:301) suggest that in evaluating quality of life there are a number of approaches utilized, including content analysis of verbal behaviour, the assessment of personal goals, the assessment of personality construct and utility/preference-based measures. They assert that the common factor is that ‘quality of life is phenomenological in nature and is specific to the individual and should therefore be defined by the individual concerned’. Increasingly, health-related quality of life outcomes are measuring both objective and subjective dimensions of life. Bhatia et al., (2002) view HRQOL as a multidimensional, self-perceived construct that includes physical functioning and activity, psychological adjustment, social functioning and relationships and overall sense of well-being.

Sredl (2004) identified six major areas of HRQOL – physical, psychological, personality, environmental, social and future orientation; each of these areas contains sub-constructs. Bekesi et al., (2011) measured the health-related quality of life changes of children and adolescents living with chronic disease (JIA, cancer and diabetes) before and after participation in a therapeutic recreation camping programme. Two hundred and fifteen children with a
chronic disease were surveyed (mean age 13.2 years) using the Kidscreen questionnaire. They found that the use of therapeutic recreation had a positive impact on the health-related quality of life of the young people, and in particular their self-perception.

Bowling (2005:9) suggests that the evidence of many writers is that ‘positive psychological outlook and emotional well-being, having good physical and mental health and the physical ability to do the things they want to do, having good relationships with friends and family, participating in social activities and recreation, living in a safe neighbourhood with good facilities and services, having enough money and being independent’ are how many people define their perception of quality of life (Farquhar 1995; Bowling et al., 2002; Bowling et al., 2003). The World Health Organization (2001) International Classification of Function in determining health-related quality of life emphasize the need for more information on the role of ‘personal factors’ and involvement in society. Campling and Sharpe (2006:171) discuss the importance of getting ‘the best from today’. They are of the view that ‘improving the quality of life today is a worthwhile aim. Making today better can also alleviate fears from tomorrow.’

Quality of life is a concept incorporating physical, mental and social aspects of life, in many cases related to health. This is known as health-related quality of life (HRQOL), which can be used for comparative purposes, to measure disease activity, to measure functional status, and to assess and determine perceived health status. It can be a basis for therapeutic decisions, for periodic update and to provide feedback; it can also be an important outcome measure in clinical trials (Testa & Simonson, 1996). Farquhar (1995) reviewed the definitions of health-related quality of life. The definitions were inconsistent and diverse; some came from a macro-level approach (societal, objective), others came from a micro-perspective (individual, subjective). The sociological perspective emphasized functionalism while the psychological perspective emphasized subjective well-being. Until recent years the concept has been under-used when assessing adolescents (Bollinger et al., 2002).
A growing body of knowledge indicates that physical activity promotes health and has a strong impact on sociological and psychological health and subjective well-being with a resultant positive impact on quality of life (Rejeski & Mihalko, 2001; McAuley & Elavsky, 2006; Berger & Tobar, 2007). This has significant implications for people with a chronic condition that impairs mobility. However, scholarly debate continues in the health promotion literature. Rejeski and Mihalko (2001), recognizing the significance of 'positive health', suggest that the definition of quality of life lacks precision until the precise relationship between physical activity and quality of life can be accurately determined.

Kleinman (1988:12), discussing the concept of illness and its meaning to the individual and the separate entity of the disease process, suggests that measuring health-related quality of life is a way of ‘translating the experience of illness’ into a quantifiable outcome. The inclusion of quality of life in the definition of health provides a more comprehensive and more encompassing definition of health. This has economic and political implications beyond the remit of this piece of work; suffice to say that patients’ growing expectations of standards of care and the reality of governments’ cost containment of care and managed care have implications for the definition of health - most likely a more limited and narrow definition with the potential for greater inequalities in child health provision (Schor, 1998).

Patric and Chiang (2000) suggest that health-related quality of life refers to those aspects of life and activities that are affected by health conditions or health services. Huebner et al. (2004:4) are of the view that health-related quality of life ‘exists at both the individual and community levels, each with its own components.’ It is clear from the literature that the concept of quality of life is multidimensional and complex. Assessing quality of life also needs to be multidimensional, otherwise only a partial ‘picture’ of the situation is provided (Ware, 1984; Stewart & Ware, 1992). Discussing adolescents in particular, Heubner et al., (2004) suggest that the inclusion of perceived quality of life
measures in national and international adolescent health databases would be very useful and could provide data to assist with improving service provision and the health and well-being of adolescents.

2.3.1 Health-Related Quality of Life (HRQOL) in Young People

Is health-related quality of life different in young people than in adults? This is a question that has exercised many commentators on the subject. The lives of children and young people are different to those of adults. The interests of children, the type of activities and social networking they engage in are different to adults. Children’s lives are affected also by the stage of growth and development across the spectrum of childhood and often proxies, usually parents, assess their child’s quality of life in the early years. The literature suggests that parents’ views and children’s views do not always concur (Tucker, 2000a; 2000b).

Advances in paediatric oncology and neonatal intensive care treatment provided an impetus and rationale for measuring quality of life in children. Lansky et al., (1985, 1987) provided early work on health-related quality of life measures. These simple measures had a strong emphasis on the clinical outcomes of morbidity and mortality and are still in use today. Work by Eisen et al. (1979), Stein (1990) and Landgraf (1996) generated generic instruments for population surveys of children. It was also common practice to measure the quality of life of children with different chronic diseases as one cohort and compare them with healthy peers (Eiser & Morse, 2001 a; 2001c; 2001d).

Who do children compare themselves with? This issue continues to create discussion. Continuing to evolve, the measures were becoming more sensitive and more refined, and new disease-specific assessments of young people with chronic diseases emerged. Acknowledging that survival rates from what would have been considered fatal conditions are improving, the residual morbidity from these conditions can include disability, emotional and learning problems in children (Newacheck & Halfon, 1998).
Changes in the epidemiology of childhood disease indicate that ‘new diseases’ are becoming more prevalent, for example diabetes (WHO 2007), asthma (Vollmer, et al., 1998), obesity (Gortmaker et al., 1987) and hyperactivity disorders (Swanson, et al., 1995). Greater consideration will need to be given to these newer areas. The views of the individual child, their level of confidence and their self-esteem can also impact on their quality of life. As in adults, assessing health-related quality of life in children is complex.

Focusing on the adolescent population, other pertinent aspects of quality of life are being integrated into tools. These may include school achievement, self-esteem and family support (Starfield et al., 1993). Healthcare providers typically find the objective and functional measures very useful while parents and young people consider the subjective measures to be intrinsic and of high value (Guyatt, 1997).

Rechner (1990) noted in a phenomenological study that the young people endeavoured to be positive and to adapt to the situation in order to maintain their normal lives throughout the illness experience and try to get on with life. These findings have been supported in the literature by Weekes and Kagan (1994) and Woodgate (2005). Woodgate (2005:8) noted in a study with adolescents with cancer and the impact that it had on their lives, that they described themselves as ‘still being pretty much the same.’

Parry and Chesler (2005) commented that adolescents with cancer used a number of coping strategies to manage their adverse circumstances. These included courage, resilience and hopefulness. However, uncertainty about the future and disruption to their normal lives (being normal) could interfere with their coping mechanisms and impact on their quality of life (Zebrack & Chesler, 2002; Santacroce & Lee, 2006). Cognitive predictive coping, such as being optimistic and maintaining a positive perspective about the course of an illness, have a strong relationship with HRQOL among early survivors of cancer. This
positive dispositive outlook can assist these young people to ‘self-sustain’ and cope with adversity. Tsakogia et al., (2011) reported similar findings in their work with adults with chronic musculoskeletal pain.

Bowling (2005) suggests that life is not only about surviving but it is important to be able to thrive. Therapies need to be assessed on the basis of their outcomes, how worthwhile they are and how they impact on the patient’s quality of life. The overarching consideration is the therapeutic value of the therapy and the child’s/young person’s best interests. Research assessing the quality of life of young people needs to be done sensitively and only in their best interests. This can lead to discussion and debate with issues such as ‘whom should we ask?’ Should the parents be asked to proxy report on the young people’s quality of life or should the young people self-report? Age and acuity of illness can impact on the validity of the child’s self-reporting assessment (Eiser, 1995; Rosenbaum et al., 1990). However, using parental proxies may result in other limitations including the young person’s subjective perspective, being excluded, parental anxiety and their level of adjustment to the situation impacting on the assessment of quality of life (Dahlquist, et al., 1994; Thompson, et al., 1992). Other writers (such as Achenbach, et al., 1987; Canning, et al., 1992; Hinds, 1990) suggest that multiple informant assessments of quality of life provide different perspectives, including the young person’s perspective, but this pathway could provide a more complete assessment of the situation. Kazak, Segal-Andrews and Johnson (1995) support this view that childhood illness impacts on all members of the family unit.

Jacobson et al., (1997a, 1997b, 1997c) discuss the importance of longitudinal studies assessing the quality of life of young people with a chronic disease. They make the point that quality of life can evolve during the life course and the continuum of the chronic disease, leading to significant adjustments or transient reactions occurring at different intervals of this disease trajectory. Cross-sectional studies may not have the capacity or sensitivity to capture these
changes or developments that can occur on the continuum of the trajectory of the disease; however, they do present a moment in time.

2.3.2 Health-Related Quality of Life – Measurement Issues

Measuring health-related quality of life can provide information about a person's health status and global well-being. It can assist in assessing the impact of treatment modalities and may provide information on the psychological impact of the condition. If the instrument used is a sensitive tool it may pick up more subtle risk factors or problems. It can also generate new knowledge about the condition, leading to potential changes in treatment and changes in the provision of services. Acquired knowledge can be a basis for future development of diagnostic therapies. It can also be a basis for healthcare systems to adapt their systems to provide more appropriate care, and the provision of practical initiatives, identifying patients’ needs with the purpose of supporting patient empowerment.

Chronic diseases have wide ranging effects, not only on the patients affected directly by the condition but also on their families. This can have a resultant impact not only on individual quality of life but also on the quality of life of the family unit of the person with the chronic disease. There can be reciprocal effects of chronic conditions and adolescent development (WHO, 2007; Simon, 2002). Treatment and side effects of treatment can also impact on growth and development (WHO, 2007).

Tucker (2006:333) suggests that ‘the impact of these disorders goes far beyond the physical symptoms and functional disability, and impacts on development, mental health, behaviour, self-esteem, the development of normal social roles (peer and family-related, school and work-related, and family functioning).’ While it is important to retain the conventional measures of physical functioning, however, a broader more comprehensive picture of the young person’s life can be gained by assessing their health related quality of life including the impact of treatments and procedures (Hatziagorou et al., 2002). Sredl (2004)
acknowledges that all of the aspects of a person’s life impact on the delicate balance of their quality of life.

Until relatively recently this area of assessing the quality of life of young people had been neglected. Adult quality of life assessment tools have been available for several decades; however, this has not been the case for children and adolescents. The appropriateness of using adult measures with children has raised concerns in the literature (Eiser & Morse, 2001a). In general, the lives and activities of young people differ greatly from those of adults. Adult generic measures to assess the quality of life of young people need consideration in relation to issues such as appropriateness of domains and items, developmental level, and the children’s cognitive skills. Utilizing adult disease-specific instruments with children, differences in aetiology and management of the disease also need to be considered (Levi & Drotar, 1998). Many commentators suggest it is important that the questionnaires/instruments used to measure the health-related quality of life of children should be developed specifically for young people (Levi & Drotar, 1998). These should be influenced by theories of child and adolescent development and theories from social psychology and sociology, and validated by an age-appropriate population (Eiser & Morse, 2001a; 2001b; 2001c; 2001d; Tucker, 2006).

Adolescence is a time of transition and the lives of adolescents are in a state of flux and change. These changes are taking place in their physical and mental development (Westbrook & Stein, 1994; Newacheck & Stoddard, 1994; Oldehinkel et al., 2004). Adapting to new environments, for example changing schools or moving from second-level to third-level education, can often present challenges for young people with and without a chronic health condition. Coping with a chronic disease in adolescence can be problematic and can impact on many facets of life, including academic performance (Shaw et al., 2006). Bullinger and Ravens-Sieberer (1995) suggested that there were few studies relating to the quality of life of young people with a chronic disease; empirical
work was scarce with few sufficiently sensitive instruments to assess health-related quality of life in children.

Duffy et al., (1993) compared the level of agreement of young people with JIA and their parents for physical and psychosocial function. It was noted from the questionnaire results that there was a high level of consensus on physical items. However, on psychosocial items the level of agreement was significantly reduced. This has implications for treatment regimens, communications and clinical decision making.

The health-related quality of life of adolescents continues to be under-researched. Also, there continues to be a need to assess cross-cultural HRQOL on a high psychometric level (Ravens-Sieberer et al., 2005).

The literature suggests that the terms quality of life and health-related quality of life continue to be defined by commentators. The continuing expansion of domains of the concept is increasing its dimensions and complexity, adding to its holistic and global approach. However, it is also creating uncertainty and difficulty in its measurement. Bandura (1997) refers to the role of self-efficacy and its implications in coping with chronic disease. Self-efficacy concerns itself with the personal judgment and belief in one’s own capabilities. It is strongly associated with personal mastery, vicarious experiences, verbal persuasion and physiological feedback.

Bandura (1977) suggests that this self-confidence predicts that the more confidence a person has in their own capabilities of coping with and controlling threatening events, the more resilient the person will be to traumatic events, experiences or life stressors. The age, stage of development, psychological and sociological characteristics of a person will impact on how they cope with a stressor and undoubtedly on their quality of life.
A range of measures are available to assess health-related quality of life. These instruments can be generic or condition-specific. Generic instruments can be used across a range of conditions while condition-specific tools are scored to reflect the problems associated with the specific condition. Scott (2000:1) suggests that 'there are trade-offs between simplicity and sensitivity and between using familiar and unusual instruments'. He is also of the view that it is appropriate in a clinical trial to include both a disease-specific and a generic tool to assess health status. These tools assess disease activity/progression and functional ability. If these tools are used on a longitudinal basis they can provide a useful comparative basis for treatment, assessment and review. Kelley-Gillespie (2009:262) has identified a number of issues with existing quality of life measurement instruments, including:

‘The lack of consensual quality of life definitions or constructs and domains being measured, the lack of rationale for using a particular measurement scale, the lack of use of summary measures, and the lack of in-depth investigations into client perceptions of relevant importance of various components of quality of life.’

Kelley-Gillespie also debated whether there were accurate health-related quality of life comparisons between populations (e.g. disabled adults, older adults, and children) and methods (e.g. interview, observation, self-administered survey).

The most commonly used condition-specific tools for adults with rheumatoid arthritis include the Health Assessment Questionnaire (HAQ), the Arthritis Impact Measurement Scale (AIMS) (Meenan 1980) and the McMaster-Toronto Arthritis Patient Preference Questionnaire (MACTAR) (Tugwell et al., 1990). The most widely used generic instrument is the SF-36. As a shorter but valid alternative to the SF-36 the SF-12 has been developed as a multi-purpose generic measure of health assessment. The SF-12 continues to gain considerable interest (Ware, Kosinski & Keller, 1995).
The Health Assessment Questionnaire includes questions addressing functional status, physical discomfort, psychological well-being, treatment side effects and economic impact. These tools aimed to integrate the broad WHO concept of health and address specific areas pertinent to patients with a rheumatic condition. Tucker (2006) is of the view that the ideal instrument should be user-friendly and straightforward to use and score. It should be applicable to a wide age range and to a diverse ethnic population. The instrument ideally would need to be able to measure physical function and quality of life. It should also have discriminative ability and its reliability and validity should be apparent.

2.4 Disease-Specific Questionnaires for Juvenile Arthritis

It was in the late 1980s that the first disease-specific instrument for juvenile rheumatoid arthritis was developed to assess the impact of the condition. This was known as the Childhood Arthritis Impact Measurement Scales (CHAIMS) (Coulton et al., 1987) and was a modified version of the Adult Arthritis Impact Measurement Scales. There is only one study published on the evaluation of this instrument. Several scales are not applicable to children under six years of age and although the scale to measure pain demonstrated good validity and reliability, other dimensions of the instrument were not considered to be sufficiently sensitive.

2.4.1 The Childhood Health Assessment Questionnaire (CHAQ)

Published in 1994 and comprising of two indices, disability and discomfort, the Childhood Health Assessment Questionnaire (Singh et al., 1994) was developed from the HAQ. The CHAQ has demonstrated excellent reliability and validity, has been translated into a number of languages and continues to maintain its reliability and validity (Singh 2001). This instrument does not attempt to measure well-being, psychosocial function or overall health; notwithstanding this, the instrument is considered to be an excellent tool within its range. Lam et al., (2004) suggest that the revised version of the CHAQ is more sensitive and less likely to have a ceiling effect, particularly with patients with oligoarthritis,
where it had been noted that there was a tendency of clustering at the normal end of the scale. However, Tucker (2006) suggests that the CHAQ may not be sufficiently sensitive to capture information relating to clinical change that may occur in patients with JIA, or growth delay or growth disturbances.

**2.4.2 Juvenile Arthritis Functional Assessment Scale and Report**

The Juvenile Arthritis Functional Assessment Scale (JAFAS) (Lovell et al., 1989) requires a health professional to observe a child carry out 10 physical tasks. Tucker (2006) consider the JAFAS to be a good instrument. However, the need for a trained observer and standardized equipment are limiting factors. The Juvenile Arthritis Functional Assessment Report (JAFAR) is an instrument completed by the patient or a parent. Both of these tools were developed from the AIMS and the HAQ and the McMaster Health Index Questionnaire (Chambers et al., 1982). Demonstrating excellent reliability and validity, the tool is used to assess the patient perform physical tasks. Tucker (2006) indicates, however, it can only be used with children over the age of seven.

**2.4.3 Juvenile Arthritis Self-Report Index**

This is a very detailed and comprehensive tool developed from the MACTAR. It is aimed at identifying physical tasks that are problematic, indicating the need for rehabilitation. These problematic areas are assessed and further evaluated sequentially. This instrument can only be used with children over the age of eight. Completion time is lengthy due to detail and exactness (Wright et al., 1996).

**2.4.4 Juvenile Arthritis Quality of Life Questionnaire**

This instrument (JAQQ) (Duffy et al., 1997) not only measures physical function but also includes psychosocial quality of life and can be administered to all age groups. Currently the unique patient scoring system makes comparisons between groups problematic. The measure has demonstrated excellent validity and responsiveness.


2.4.5 Childhood Arthritis Health Profile

The Childhood Arthritis Health Profile (CAHP) aims to assess both physical and psychosocial function. It includes both generic and condition-specific scales. The generic scales of the CAHP are from the Child Health Questionnaire and comparisons can be made across other populations of children. This is a detailed, comprehensive instrument with a lengthy completion time more likely to suit longitudinal research studies than the clinical setting. It measures the impact on function of physical function, pain, emotional and behavioral function, mental health, self-esteem and morning, the specific impact of JIA on school and social function, family function and the impact of JIA on parent emotions and their time for themselves.

2.4.6 Paediatric Rheumatology International Trials Organization Clinical Assessment

The Paediatric Rheumatology International Trials Organization developed a set of core outcome variables. These include patient or parent global assessment of disease impact, physician’s global assessment of activity, number of active and limited joints, an index of inflammation and a functional assessment by parents. This is a clinical functional assessment tool which could be used in conjunction with a quality of life tool to provide a more comprehensive assessment of the young person’s current situation.

2.4.7 The Pediatric Quality of Life Inventory (PedsQL)

This 23-item modular instrument was developed to measure the quality of life of young people aged 2-18 years. It comprises four scales which measure physical, emotional, social and school functioning. The generic core measure can be integrated with PedsQL disease specific-specific modules. The disease specific module has five scales which measures pain, daily activities, treatment, worry and communication. The instruments are available as child-self report and parent proxy-report. These instruments have demonstrated reliability, validity and responsiveness in both the core instrument (PedsQL 4.0 Generic
core scales and the rheumatology specific module (PedsQL 3.0 Rheumatology module) (Varni et al., 2002).

2.4.8 The DISABKIDS Health-Related Quality of Life Measure

The DISABKIDS questionnaire is a European quality of life assessment tool devised within the Fifth Framework Research Programme quality of life and management of living resources. It aims to assess the psychosocial impact of chronic health conditions on quality of life. Bullinger (2002:198) defines quality of life as ‘subjective perception of health in physical, emotional, mental, social and functional domains. It is represented in terms of well being as well as functioning’.

The DISABKIDS health-related quality of life chronic/generic questionnaire has six dimensions: independence, emotion, social inclusion, social exclusion, limitation and treatment. These have evolved from the three domains of mental, social and physical HRQOL as conceptualized by the World Health Organization. This questionnaire embeds generic questions, which can be used for comparative purposes with healthy peers.

The DISABKIDS condition-specific instruments evaluate the health-related quality of life of children and adolescents using condition-specific questionnaires for the following conditions: asthma, arthritis, dermatitis, diabetes, cerebral palsy, cystic fibrosis and epilepsy (Peterson et al., 2005; Simeoni et al., 2007; Sandeberg et al., 2010). The arthritis questionnaire has been developed to supplement the DISABKIDS chronic/generic questionnaire. It focuses on physical symptoms and limitations. It has two main domains, ‘impact’ and ‘understanding’.

The impact scale is divided into two sub-domains, pain and limitations. The ‘impact’ scale evaluates the physical symptoms and the effects of the disease on activities of daily living. These include pain and functional and social limitations that the young person may have as a result of the disease.
The ‘understanding’ scale has been developed to assess the social impact of the disease on the person. It evaluates social inclusion, social exclusion, and empathy of peers and teachers.

The DISABKIDS questionnaire is the instrument being used in this project. Its user-friendly modular format using a ‘Likert’ scale and the embedded generic questions allows it to be used with a group of healthy peers. The domains of the DISABKIDS tool are independence, physical limitation, emotion, social inclusion, social exclusion, and treatment.

The domains of the generic component are physical well-being, psychological well-being, moods and emotions, self-perception, autonomy, parent relationship and home life, peers and social support, school environment, bullying, and financial resources. The domains in these tools were thought to be more comprehensive than in other tools considered.

This questionnaire is also available in several languages, potentially enabling comparative work in similar small populations in Europe. This arthritis disease-specific questionnaire had embedded in it both generic and condition-specific health-related quality of life questions to be given to the group with JIA. An advantage of this tool was that there was also a separate short-form generic questionnaire that could be given to a comparator group without arthritis.

Emery (2004) suggests that while some instruments have been standardized and validated across populations they are not necessarily sufficiently sensitive to small fluctuations in function or variations in specific joints’ range of movement. Emery also makes the point that new technologies and therapies can provide excellent information for studies; these include gait analysis laboratories, although currently this is expensive and inaccessible for many. In the future it is possible that a multidisciplinary approach to assessing health-related quality of life could provide a more comprehensive view of the young person’s functional abilities and quality of life.
When assessing patients with JIA a global approach, including the effects of persistent damage or non-reversible damage, needs to be considered. Other issues that require assessment include reversible active inflammation, pain and functional status (Palmisani, 2006). The historical pathway of outcome measures for juvenile arthritis first commenced in the 1980s and concentrated on disease activity. During the 1990s the focus changed to the patient’s functional ability. Further development took place during the 2000s to include disease activity, functional ability and health-related quality of life. There is, however, no validated tool for the definition of remission of the condition (Zulian, 2003). Janse et al., (2005) note that there can be substantial discrepancies in parents’ and physicians’ perceptions of the health, well-being and quality of life of children with JIA; parents were more likely to provide a lower rating.

Duffy (2004) acknowledges that there is a need for greater assessment of the health status, functional status, quality of life and socioeconomic status of individuals with JIA in order to provide a more comprehensive and complete assessment of young people with the disease. Duffy also indicates that there continues to be problems associated with JIA that are not being adequately addressed, for example high rates of arthroplasty as a result of joint damage, and increasing problems of osteopenia and osteoporosis, which can have enduring and long-term effects.

Sawyer et al., (2005), in a 12-month prospective study, compared health-related quality of life (HRQOL), coping strategies and experience of pain. Despite many of the young people’s illnesses being stable and relatively mild, their HRQOL was found to be significantly worse than their peers. The reasons for these differences can be multifactorial and individual. For example, communications with adolescents can be problematic for parents and consequently their pain may go unrecognized. Sawyer suggests that young people may internalize their problems and only externalize them at a remove from the family home.
Recommendations for clinical practice include the implementation of new approaches for early identification of problems before they become more severe. Sawyer also suggests that it is important to be aware that reports from either the young people or the parents may not accurately reflect the perceptions of the other.

Service delivery, its timeliness, its effectiveness and efficiency is another factor associated with health-related quality of life (Guyatt, 1997; Hack, 1999; Chernoff et al., 2002; Law et al., 2002; BSPAR 2009; ARMA 2010).

The issue of capturing depth of information when using questionnaires as a tool arises also. Wolfe and Sirios (2008), using both a generic and a condition-specific questionnaire to explore the quality of life experiences of adults (n=282) with inflammatory bowel disease (IBD), noted that a number of dimensions of HRQOL were not captured using these measures including cognitive, self-regulation and practical dimensions. Asking the participants the open-ended question ‘How has IBD affected your daily activities?’ provided a rich source of data. The focus of the research was on the daily activities of the person and the impact of the disease on these daily activities rather than the overall impact of the disease. This study provides support to the argument for the use of both a questionnaire and an interview to provide the richest range of data.

2.5 Summary

In conclusion, health and quality of life are intrinsically linked. Health-related quality of life is the assessment of quality of life within the context of clinical medicine and clinical research and describes an approach used to assess development and functioning (Fayers & Machin, 2002). This approach focuses on the patients’ unique perspectives of their own health viewed from a number of angles including physical, psychological and social domains (Sawyer, et al., 2004). Often these multidimensional terms quality of life and health-related quality of life are used interchangeably, frequently causing confusion. It is
recognized that healthcare providers may be able to influence certain limited aspects of a person's quality of life related but this may not extend to influence their more global perspective of quality of life (Tucker, 2000a).

Parts 1.1-1.3 of this chapter have outlined the available instruments for application with young people with juvenile idiopathic arthritis. Many of them have been developed to focus on physical function only, while others attempt to measure quality of life in addition to physical function. Some of the instruments, for example the CAHP, are very detailed. These can provide excellent data, but a limitation is they can take a long time to complete, which may cause some difficulty in administration and response rate. Using a global and holistic approach, more reliable and sensitive measures are needed to evaluate the health and quality of life of patients with JIA, which is such an unpredictable disease.

Generic measures may provide information on global well-being but may not be sensitive to a particular population. In an era of electronic innovations, including interactive computer games, some of these instruments discussed and those of the future will doubtless be further refined to be more ‘user-friendly’ to young people, to capture the depth of information, the particular perspective and the interest and response of young people.
2.6 What is Juvenile Idiopathic Arthritis?

Juvenile idiopathic arthritis is the umbrella term for a group of chronic inflammatory arthritis conditions that occur in young people. The aetiology is unknown and the disease is characterized by exacerbations (‘flare-ups’) and remissions. The disease is variable and can be difficult to diagnose. The diagnosis criteria include the condition commencing before the young person is 16 years of age, the episode lasting for more than six weeks and where known causes have been excluded (Cassidy & Petty, 2011; Davidson, 2000). Juvenile’ is defined as a young person under the age of 16. ‘Idiopathic’ means the cause of the illness remains unknown, and ‘arthritis’ ‘indicates an inflammatory process taking place in joints, characterized by painful, warm, swollen and stiff joints’ (Beresford 2011:162).

The disease can persist into adulthood and can have significant morbidity (Selvaag et al., 2006; Van Rossum et al., 2003; Zak et al., 2000; Minden et al., 2002; Bowyer et al., 2003; Packham & Hall, 2002). This section aims to provide an overview of the complexity of the signs and symptoms of juvenile idiopathic arthritis. It does not aim to provide a medical textbook account, but rather to indicate the opportunities for multi-strategy health promotion strategies in a variety of settings.

Juvenile idiopathic arthritis (JIA) is a heterogeneous autoimmune disease characterized by inflammation in one or more joints. Many commentators agree that two thirds of young people with the disease in childhood will not continue to have the condition as adults however there is a significant number who will have it during their adult years (Gowdie & Tse 2012; Cassidy et al., 2011; Oen et al., 2002; Gare & Fasth, 1995; Cooper 1994). Consequently, early diagnosis and referral to a specialist centre are critical to minimize joint, growth and other potential problems (BSPAR 2010).

Emery (2004:1382) states that ‘early identification of children with rheumatic disease and aggressive intervention with a combined medical, rehabilitation,
Typical symptoms of juvenile idiopathic arthritis (JIA) include morning stiffness and stiffness following a period of inactivity. Acute and chronic pain, swollen joints, limited mobility, functional impairment, fatigue, lack of appetite and generally feeling unwell are all features of the condition. Young people with JIA can present also with non-specific musculoskeletal signs and symptoms, which can include painless joint inflammation. Laboratory results may also be within the normal range (Junnila & Cartwright, 2006). Len et al., (2006) acknowledge that the initial signs and symptoms of the disease can be non-specific. For example, there is relatively low prevalence, there is an absence of pathognomonic clinical signs, clinical manifestations can be similar to orthopaedic, infectious and malignant diseases and some presentations are atypical.

The incidence of the disease is approximately 1 in 10,000, and the prevalence is 1 in 1000 (Friswell 2004; Ravelli & Martini 2007; Beresford 2011). Norwegian statistics suggest an annual incidence of 14 and a prevalence of 130 per 100,000 children less than 16 years of age (Flatø 1999). Manners and Bower (2002) indicated that prevalence values varied considerably with 7 to 401 per 100,000 children in their review between 1966 and 1998 of 34 epidemiological studies. Other more recent work suggests the prevalence values vary between 16 and 150 per 100,000 young people (Ravelli & Martini, 2007). Andersson-Gare (1999) suggests that 60% of children affected with JIA are girls. Diagnostic difficulties, small sample sizes, ongoing definition differences in disease criteria, methods of data collection and healthcare resources could go some way to explaining these differences in values by commentators (Modesto et al., 2010).

Currently there is no convincing evidence of what activates the chronic inflammatory process. However, genetic predisposition, environmental factors, injuries, surgical procedures and the condition being triggered as a result of a
viral infection are considered potential risk factors (Southwood, 2006; Modesto et al., 2010). There are two peak ages of onset: between 1 and 3 years of age and 8-10 years of age (Cassidy et al., 2011). Diagnosis can be complex, and is made following a detailed history, clinical examination and exclusion of other disorders. Symptoms can be subtle and similar to other diseases. Laboratory tests, while helpful, are rarely diagnostic (Szer, 2006). There is an increasing trend for the use of ultrasound imaging for assessment of joints and to monitor disease activity in JIA (Collado et al., 2012). Early and aggressive treatment to control and ‘switch off’ the inflammatory process is the goal of intervention (Cassidy et al 2011; Beresford, 2009; Wallace, 2010) (please see Appendix 2).

Young people with JIA tend to have morning joint stiffness and pain, or joint stiffness and pain after a period of inactivity. This is referred to as ‘gelling’ of the joints (Beresford, 2011). Joints can become stiff and swollen with a consequent loss of motion occurring in these affected joints. The joints may be warm to touch but rarely red in colour. The limited motion in the early stages of the disease is usually due to muscle spasm and the joint/s being swollen. At later stages of the disease limited motion is caused by soft tissue contractures. Young people may be stiff or in pain but they do not always state this; cognitive development may be a factor in these situations (Cassidy et al., 2011). Often children will try and cope by avoiding pain to the area; for example, if they have problems with their knees and if having difficulty coming down the stairs they may come down on their ‘bottoms’, avoiding pressure on their knee joints. Cassidy et al., (2011) refer to this as a physical expression of a pain rather than a vocal expression. Another reason for not indicating that they have pain includes habituation to the pain (Von Baeyer, 2007).

The activated inflammatory process causes the joints and tissues surrounding the joints to become swollen. As with many diseases the spectrum of the condition can range from mild to moderate to severe. If the condition is not treated or is resistant to treatment this would allow inflammation of the tissues
surrounding the joint to increase and inflammation of the joint would eventually cause the joint to be worn away and destroyed.

Duffy (2005:369) is of the view that JIA is not a benign disease. The disease often continues into adulthood. 'Most remissions occur in the first 5 years after disease onset. The probability of remission 10 years after onset approximates 30%-35%, overall, and decreases progressively thereafter'. Duffy, who has written extensively on this subject, suggests that as a result of improved therapies functional outcomes have improved significantly. However, he goes on to state 'the degree of impaired functional outcome is still unacceptable and needs to be improved. Significant joint damage is still a consequence of this disease, with relatively high rates of arthroplasty, and increasing data reveal significant problems of osteopenia and osteoporosis that are not being addressed adequately' (please see Appendix 3).
2.6.1 Types of JIA

Improving rheumatology knowledge is providing clinicians with a greater understanding of the condition, allowing them to appreciate and further refine and differentiate a number of subsets of the disease. This process continues to evolve and it is likely that further classifications will emerge (Southwood 2006). The International League of Associations for Rheumatology (ILAR) classifies JIA into seven main types: oligoarticular JIA, seropositive polyarticular JIA, seronegative polyarticular JIA, systemic-onset JIA (sJIA), enthesitis-related arthritis (ERA), psoriatic JIA (PsJIA) and undifferentiated JIA (Gowdie & Tse 2012). (please see Table 2.1). 

2.6.2 Oligoarticular JIA

This type of arthritis which is also referred to as oligoarthritis affects between one and four joints in the first six months of the disease. This is the commonest form of juvenile arthritis and mainly affects the large joints, for example, the knees, ankles, wrists and elbows. The occurrence of uveitis with oligoarthritis is 30% (Gowdie & Tse, 2012). Oligoarthritis is the most common form of JIA with frequently an insidious onset. The child may present with joint stiffness and pain, which is improved by activity and exacerbated by inactivity. Fluctuating symptoms can confound the situation for parents and professionals alike, as a child who is feeling stiff in the morning may be out playing some hours later. Children do not always complain of pain (Miller & Malleson 2006). Instead they may not use the limb/s and may stop walking or standing, especially in the morning. The child may be ‘fussy’ or uncharacteristically quiet. These symptoms are suggestive that the child is in pain (Britton, 2004).
<table>
<thead>
<tr>
<th>ILAR JIA Subtypes</th>
<th>Age, Sex, and % of Total Patients with JIA</th>
<th>Typical Joint Involvement</th>
<th>Occurrence of Uveitis</th>
<th>Other Features</th>
</tr>
</thead>
<tbody>
<tr>
<td>Oligoarticular Persistent</td>
<td>F&gt;M</td>
<td>≤ 4 joints; Large joints: knees, ankles, wrist</td>
<td>Common (30%)</td>
<td>Especially if ANA-positive</td>
</tr>
<tr>
<td>Extended</td>
<td>Early childhood 40%-50%</td>
<td>Persistent disease: never &gt;4 joints affected</td>
<td>Usually asymptomatic</td>
<td>ANA 60%-80% positive</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Extended disease: involves &gt;4 joints after first 6 months of disease</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Polya rticular (RF-negative)</td>
<td>F&gt;M</td>
<td>≥ 5 joints; Symmetric</td>
<td>Common (15%)</td>
<td>ANA 25% positive</td>
</tr>
<tr>
<td></td>
<td>2 peaks: 2-4 yrs and 6-12 yrs 20%-25%</td>
<td>Poly or oligoarticular</td>
<td></td>
<td>≥ C spine and TMJ</td>
</tr>
<tr>
<td>Polyarticular (RF-positive)</td>
<td>F&gt;M</td>
<td>Symmetric small and large joints</td>
<td>Rare (&lt;1%)</td>
<td>ANA 75% positive</td>
</tr>
<tr>
<td></td>
<td>Late childhood/early adolescence 5%</td>
<td>Erosive joint disease</td>
<td></td>
<td>Rheumatoid nodules</td>
</tr>
<tr>
<td>Systemic</td>
<td>M = F</td>
<td>Poly or oligoarticular</td>
<td>Rare (&lt;1%)</td>
<td>Daily (quotidian) fever for ≥ 2 weeks</td>
</tr>
<tr>
<td></td>
<td>Throughout childhood 5%-10%</td>
<td>Symptomatic acute Uveitis (~ 7%)</td>
<td>Evanescent rash, Lymphadenopathy</td>
<td>Enthesitis</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Hepatosplenomegaly</td>
<td>HLA-B27-positive</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Axial involvement (including sacroiliitis)</td>
<td>Axial involvement (including sacroiliitis)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Family history of HLA-B27-associated disease</td>
<td>Family history of HLA-B27-associated disease</td>
</tr>
<tr>
<td>Enthesitis-related arthritis</td>
<td>M&gt;F</td>
<td>Weight-bearing joints especially hip and intertarsal joints</td>
<td>Symptomatic acute Uveitis (~ 7%)</td>
<td>Serositis</td>
</tr>
<tr>
<td></td>
<td>Late childhood/adolescence 5%-10%</td>
<td>History of inflammatory back pain or sacroiliac joint tenderness</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Psoriatic arthritis</td>
<td>F&gt;M</td>
<td>Asymmetric or symmetric small or large joints</td>
<td>Common (10%)</td>
<td>Nail pits, onycholysis</td>
</tr>
<tr>
<td></td>
<td>2 peaks: 2-4 years and 9-11 years 5%-10%</td>
<td></td>
<td></td>
<td>Dactylitis</td>
</tr>
<tr>
<td>Undifferentiated</td>
<td>10%</td>
<td></td>
<td></td>
<td>Does not fulfil criteria for any above category or fulfils criteria for &gt;1 category</td>
</tr>
</tbody>
</table>

Table 2.1: Characteristics of JIA
Oligoarthritis has two subsets: ‘persistent oligoarthritis’ and ‘extended oligoarthritis’:

**Persistent oligoarthritis** – there are never more than four joints affected by arthritis. As the young person becomes older, there is a high incidence of this type of the disease becoming less active and becoming more quiescent, often without any residual damage to the joints (Weiss & Illowite 2005).

**Extended oligoarthritis** – the disease extends to more joints after six months of having the condition (Weiss & Illowite 2005). This occurs in approximately 50% of patients with oligoarthritis (Gowdie & Tse 2012).

A particular complication of oligoarthritis is anterior uveitis (please see section) 

The physical examination of children is very important, with emphasis on joints and eyes. Joints are examined for tenderness and warmth. Growth problems of both undergrowth and overgrowth can occur in this form of arthritis, resulting in serious joint complications

### 2.6.3 Polyarticular JIA

This type of arthritis also referred to as polyarthritis affects more than four joints. It affects one in four children with arthritis, mostly girls, and it is the second most common form of JIA. This form of arthritis is divided into two subsets: polyarthritis rheumatoid factor negative (RF-negative) and polyarthritis rheumatoid factor positive (RF-positive).

#### 2.6.3.1 Polyarthritis (rheumatoid factor negative)

This type of arthritis which involves more than 5 joints can affect children at any age. There is symmetric joint involvement. Joints to be affected include the hands, feet, hips, knees, neck, elbows, shoulders and jaw. The total percentage with polyarticular (RF-negative) is approximately 20%-25% (Gowdie & Tse 2012). Young people with this type of arthritis can appear very unwell. They can develop a high temperature (pyrexia) when a flare-up occurs and
joints will be inflamed. Approximately 25% of young people with polyarthritis rheumatoid factor negative (RF-negative) will go into remission, while 75% will continue to have an active arthritis condition (Weiss & Ilowite 2005).

### 2.6.3.2 Polyarthritis (rheumatoid factor positive)

This form of arthritis occurs in less than 1% of all children with JIA, typically it occurs in late childhood/early adolescence, with symmetric small and large joints affected (Gowdie & Tse 2012). The rheumatoid factor positive antibody is similar to that found in adults with rheumatoid arthritis. This type of arthritis affects mostly girls aged ten years or older. It can be a severe and aggressive form of arthritis and needs to be treated and monitored closely to avoid erosive damage to the joints (Weiss & Ilowite 2005).

### 2.6.4 Systemic Arthritis

As the name suggests, this form of arthritis affects the whole body and not just particular areas. It can occur throughout childhood and affects 5-10% of young people with JIA (Gowdie & Tse 2012). The young person may have swollen glands (Lymphadenopathy) and a high temperature once or twice a day for approximately 2 weeks. A blotchy pink rash (evanescent rash) may be noted on the thighs, arms or body. The axillae are a frequent site of the rash, which may be migratory (McDonagh, 2006). Tissue surrounding the heart and the lungs can also be affected by inflammation. The joints may not be swollen at this time but inflammation of the joints can occur later. It is very difficult to diagnose and can be confused with other serious diseases, such as meningitis, leukaemia and the measles (Weiss & Ilowite 2005). Consequently, parents are likely to be very anxious. As yet, there is not a unique diagnostic test, therefore it is necessary to exclude other diseases. The young person may have to undergo invasive tests and families may have a ‘worrying wait’. The young person is likely to be feeling acutely ill; however, it is frequently noted that when the diagnosis is made families are relieved that it could have been worse than they had feared (Britton 2004). The prognosis for these young people is that 60% will have a good
recovery after a few years and 20% will develop ongoing severe arthritis, while others will have ongoing cycles of remission and flare-ups (Britton, 2004).

2.6.5 Psoriatic Arthritis
This form of arthritis is associated with psoriasis. It affects 5-10% of young people with JIA. It occurs in more girls than boys. There are 2 peak ages for this type of arthritis 2-4 years and 9-11 years. Occurring symmetrically or asymmetrically, both small and large joints are affected (Gowdie & Tse 2012). The young person may have arthritis for some time before the psoriasis becomes apparent. There is a strong family history with this form of arthritis. Nail pits, onycholysis and dactylitis can occur with this type of JIA. It is also associated with inflammation of the eye, uveitis, occurring in 10% of the JIA population (Weiss & Ilowite 2005).

2.6.6 Enthesitis-Related Arthritis
Enthesitis is inflammation at the point where the tendons are attached to the bones. This form of arthritis affects the hips, knees, ankles and frequently the sacroiliac joints (where the base of the spine joins the pelvis). Often it is associated with a history of inflammatory back pain (Gowdie & Tse 2012). It can affect other joints also. The condition typically affects more boys than girls usually over the age of 8 years. This form of the disease is associated with the condition ankylosing spondylitis in adults and the HLA.B-27 genetic marker. Three out of four children with enthesitis-related arthritis carry the HLA-B27 genetic marker. However, not all of these young people will develop ankylosing spondylitis. Enthesitis-related arthritis is associated with acute anterior uveitis affecting approximately seven per cent. This condition is found less frequently than the eye condition associated with oligoarthritis. (Weiss & Ilowite 2005).

Undifferentiated Arthritis
This type of arthritis occurs in 10% of the JIA population (Gowdie & Tse 2012). Patients are placed in this category if they fulfill the criteria for more than one
sub-type of JIA or if they do not fulfill sufficient criteria for a particular sub-type of JIA (Southwood & Kimura 2006).

**Complications of JIA**

**Uveitis**

Uveitis is a serious complication in JIA. This inflammatory condition of the eye, primarily in the iris and ciliary body, can occur in oligoarthritis (30%), polyarthritis (RF-negative) (15%); polyarthritis (RF-positive) (<1%); systemic arthritis (<1%); enthesitis-related arthritis (7%) and psoriatic arthritis (10%) (Gowdie & Tse 2012). It can be related to ocular diseases such as cataracts and glaucoma where visual impairment may result, or as an isolated phenomenon unassociated with JIA (Ozdal et al., 2005; Heiligenhaus et al., 2007; Thorne et al., 2007). The onset can be insidious and painless in oligoarthritis, in contrast to enthesitis-related arthritis when the onset can be very painful. Regular ophthalmic monitoring is required to detect the condition. If the condition is identified, close ophthalmic monitoring to judge the response of treatment is required (Gowdie & Tse 2012). Regular installation of eyedrop medication can have an impact on the quality of life of the young person with this type of arthritis. The vast majority of this group of patients require methotrexate and up to 25% require biologic therapy to control the uveitis.

**Synovial Inflammation**

Prolonged synovial inflammation may lead to permanent changes in joint structures. Permanent changes may also take place in extra-articular organs/systems, for example the eye kidney (due to systemic amloidosis) (Cassidy & Petty, 2001), or may result from side effects of medications.

**Growth Abnormalities**

Growth disturbances can occur in young people with JIA. These can be linear disturbances or localized disturbances, causes are considered to be multi-factorial. Chronic inflammation and high levels of circulating proinflammatory cytokines are considered to be contributory factors (Gowdie & Tse 2012).


2.7 Principles of Management of the Disease

The goals of management are ‘to control inflammation with medications; normalize range of movement and function; maintain normal motor development and facilitate normal behavioural, psychosocial educational and vocational development’ (Emery, 2004:3).

Pharmacological Interventions

The aim of this section is to discuss in general terms the pharmacological approach to the treatment of juvenile idiopathic arthritis.

The general management principles that apply to juvenile idiopathic arthritis are to suppress the abnormal inflammatory process early, quickly and completely, and return the patient to normal physical and vocational function with structurally normal joints (Duffy 2006). The heterogeneity of the disease and its sub-groups, however, explains the need for individual tailoring of treatment. Successful management of the disease reduces inflammation and pain and facilitates the return of the young person to their normal activities, which has a direct impact on their quality of life. In more recent years, improved knowledge relating to the pathophysiology of the disease is facilitating clinical trials research to aim for more targeted therapies (Ruth & Passo 2012).

The aim is to attempt to control the condition by suppressing inflammation. If non-steroidal anti-inflammatory drugs (NSAIDs) are not effective methotrexate (MTX), a disease-modifying anti-rheumatic drug (DMARD), is now the gold standard treatment in moderate to severe forms of JIA. This medication has been used since 1980 (Giannini et al., 1997). Methotrexate, however, has been found to be less responsive in systemic JIA (Halle & Prieur, 1991; Woo et al., 2000). In recent years intra-articular and intravenous ‘pulse therapy’ corticosteroids are being used rather than the long-term high-dose use of oral corticosteroid therapy as an adjunct to DMARD therapy (Murray & Lovell, 2002).
Young people with resistant forms of JIA to a variety of pharmacological interventions, including high-dose intravenous methylprednisolone, methotrexate given parenterally, cyclophosphamide and newer medications such as anti-tumour necrosis factor-α (anti-TNFα) (Quartier et al., 2010) such as etanercept, may have more treatment options with the introduction of more experimental therapies.

Göksel et al., (2005), in a small-scale study investigating renal disease occurring in young people with JIA due to the disease or as a result of the side effects of the medications, found an increase in tubular enzymuria during the active phase of the disease. However, the study found no occurrence of permanent renal damage.

Lovell et al., (2008), in a study to evaluate the safety and efficacy of the use of etanercept treatment in juvenile idiopathic arthritis over a period of eight years. Acknowledging that this was a small study, Lovell et al., concluded that there was a sustained benefit on the patients’ signs and symptoms and these improvements were maintained as a consequence of controlling disease activity.

Glucocorticoid therapy which is a fast acting therapy continues to be used as a short-term 'bridging therapy' until conventional DMARD therapies which are slower acting therapy, impact on the disease activity, however the evidence for systemic use of steroids is such that its use has not been discussed in the ACR 2011 guidelines (Sawhney 2012).

All medications can have side effects, some very bothersome. Clear lines of communications between patients and clinicians to report adverse effects are integral to the treatment plan. Close monitoring of blood levels is required to observe for abnormal deviations. However, there can be short-term side effects of the medications such as nausea, which can cause distress and anxiety and can impact on the young person’s quality of life. Long-term side effects of some medications could also have implications for quality of life; for example, the
young person could be more susceptible to osteoporosis, which could in later years mean that they are more susceptible to fractures. Outcomes for children with rheumatologic disease are improved if managed in a multidisciplinary setting with the input of a paediatric rheumatologist (Kurtin, 2000; Cuesta et al., 2000).

Invasive procedures to administer the medications, including subcutaneous injections and venepunctures, can be painful causing distress to the young people (Shaw et al., 2006). These procedures if performed regularly may cause anticipatory anxiety in some young people, which can impact on their quality of life.

A well-known complication of JIA is growth impairment. The use of glucocorticoids has also been associated with affecting growth. Advances in new therapies, particularly anti-tumour necrosis factor (TNF), are improving in suppressing inflammatory activity but also in restoring growth velocity (Tynjälä et al., 2006). Studies into intra-articular corticosteroid injection (ICI) use in the lower limbs and evaluation of 3D-gait analysis suggests positive results especially relating to joint movement, walking velocity and pain (Broström, et al., 2004).

Alsufyani et al. (2004) conclude that for children with JIA who are unable to take oral methotrexate due to intolerance of the medication, greater tolerance and effectiveness of the medication has been noted when it is given by the subcutaneous route. This route has the potential advantage of improved absorption and high drug bioavailability. This route of administering the medication is more invasive to the young person and can cause pain and anxiety.

There is some evidence in adult rheumatoid arthritis to suggest that aggressive treatment in the early stages of the disease is more effective than later in the course of the disease (Moreland & Bridges, 2001). In recent years paediatric
rheumatologists are implementing more aggressive treatment regimens in the early course of the disease.

Kileen and Gardner-Medwin (2006), reviewing the literature on the use of folinic acid to counteract the side effects of methotrexate, identified nine papers in their search but only three relevant papers. The review of the papers considers methotrexate to be generally well tolerated in young people with JIA and folinic acid may counteract the adverse side effects of MTX, but the evidence was insufficient to conclude that folinic acid impairs the effect of methotrexate.

The use of adalimumab therapy in young people with persistent uveitis is indicating a good response in situations where there had been a poor response to conventional therapy. Vazquez-Cobian et al., (2006) indicate that adalimumab may be a useful therapy in young people with uveitis associated with juvenile idiopathic arthritis.

Pharmacological decision making can be assisted by the use of treatment algorithms. The algorithm guidelines can be refined further to provide guidance for early and established disease.

Ethical considerations address the safety and efficacy of new therapeutic approaches. The use of new treatment modalities needs constant monitoring and vigilance and the early identification of side effects. This monitoring needs to be maintained into adulthood. Prospective data collection in a central registry and appropriate supervision of the use of new therapies are important elements in the care of patients with JIA (Horneff et al., 2011). Effective education of patients, families and practitioners and an integrated approach to care is of paramount importance (Gerhardt et al., 2011).

2.8 Pain Assessment and Management

The aim of this section is to discuss pain associated with juvenile idiopathic arthritis, assessment of pain in young people and its management. Young
people with JIA can experience pain, stiffness, lack of energy/stamina and altered body image. These issues can impact on psychosocial development, and as a consequence interaction with peers. Managing a medication regimen at home and at school can also be problematic for a teenager. (ARMA 2010).

The common definition of pain is ‘an unpleasant sensory and emotional experience, associated with actual or potential tissue damage or described in terms of such damage’ (International Association for the Study of Pain, 1979).

The subjective nature of pain and the child’s development status play a role in their response to pain. Significant fatigue and lack of energy are common symptoms of polyarticular or systemic arthritis. This may be due to ‘a flare-up’ or poor control of the condition. Quality of life can be affected by fatigue and pain (Ringold 2013). Managing pain appropriately often improves fatigue, consequently improving the overall situation and perceived quality of life of the young person.

Young people with JIA will tend to keep their joints in the most comfortable position. Frequently this will be a position of flexion for the joint and consequently extensor muscles can become weak and flexor muscles can contract. Pain, stiffness and fatigue can contribute to the young person being less active than their peers. Reduced mobility can lead to deficits in flexibility, exercise capacity and cardiovascular reserves (Emery et al., 1995; Klepper, 2003; Klepper 2011).

The use of electronic technology to assess pain and disability in adults and paediatric patients is increasing (Lootens & Rapoff, 2011). This technology is becoming more ‘user-friendly’ for young people as a method of ongoing pain assessment to improve pain management. Self-report measures such as ‘e-ouch electronic pain diary’, and other electronic versions of pain measurements, may be feasible for applications for personal electronic devices and ‘Smartphones’ (Lootens & Rapoff, 2011).
Sleep disorders and fatigue are frequently associated with musculoskeletal conditions (Drewes et al., 1998; Labyak, 2003; Ringold et al., 2013). They are also associated with pain; however, the association can be complex. Pain can exacerbate the sleep disorder and the sleep disorder can increase the pain. Passarelli et al., (2006), investigating nocturnal sleep disruption in children with polyarticular juvenile rheumatoid arthritis, reported that pain is related to fragmented sleep in young people with juvenile arthritis. However they indicated a number of limitations of the study including the small number of participants, the absence of assessment of participants' iron levels – low iron levels can affect sleep - and the recognition that some medications can provoke sleep disorders, including nonsteroidal anti-inflammatory drugs.

Thastum et al., (2001), reporting on a small-scale study, suggested that young people with juvenile arthritis may differ in their coping strategies and their responses to experimental pain from the control group. Grag et al., (1996) suggested that psychosocial factors could contribute significantly in the perception of pain of young people with juvenile arthritis.

Janse et al., (2005) suggests that there can be perception differences between parents and physicians also, in their assessment of pain in children with chronic diseases, with physicians underestimating the pain. This has implications for the young people and the doctor/parent-patient relationship. Children may have difficulty communicating the level of intensity of their pain or discomfort or may be dismissive of it. It is important that pain and discomfort is treated. Accordingly, pain needs to be assessed using a variety of approaches (Sawyer et al., 2004). Britto et al., (2004) in a study considering the health care preferences and priorities of adolescent with chronic illnesses identified minimizing pain as an issue. Procedural pain for example, venepuncture, it was suggested could be reduced by improving the technical competence of health care providers.

Sherry et al., (1990) note that even with active disease young people with juvenile arthritis may not talk about their pain. It is speculated that inflammation can be insidious, and as a consequence the young people can

Major factors in the development of functional disability in the juvenile arthritis are loss of motion and decreased physical function (Lemanek et al., 2001). Chronic pain can lead to attention deficits which can have implications for meeting education targets (Eccleston, 1994, 1995; Asmundson et al., 1997; Eccleston et al., 1997; Crombez et al., 1998).

The social importance of play, including belonging and social participation, is important for all children but take on an added significance when the young person has a chronic disease. Attendance at appointments, treatments and exercise programmes can reduce leisure and play time. Absences from school and school work will, of necessity, have to be made up. This can be an incursion into leisure time and can have significant social implications for the young person (Shaw 1999). Adaptation of situations to avoid the young person being ‘left out’ and feeling isolated from peers is important for the young person’s psychosocial well-being.

Hackett (2003), in her work with young people with arthritis, suggests that the impact of the disease, including fatigue, can result in reduced physical activity. As a consequence, this can create situations of social isolation and feelings of sadness and of being different. It can also be a factor in physical deconditioning and create a vicious circle effect. Ringold et al.,’s (2012) more recent work supports Hackett’s views.

Kyngäs and Rissanen (2001) carried out a study to identify the issues that predict adolescents adhering to their treatment regimen. Questionnaires were completed by groups of 300 young people with asthma, epilepsy, juvenile rheumatoid arthritis and insulin-dependent diabetes. Twelve hundred young people were identified and a final response rate of 88% was obtained. Support was the key predictor of compliance amongst all of these groups of young
people with different chronic diseases. Support could come from a number of people including parents, nurses, physicians and friends. Nurses were seen as the most significant people in the provision of support. Other factors that predicted good compliance were motivation, energy and willpower. Kyngäs and Rissanen also noted that a sense of partnership with the healthcare team, and being able to negotiate their treatment regimen was also seen as important.

**Non-Pharmacological Interventions**

Management of young people with JIA is normally on an outpatient basis. It requires a co-ordinated approach by an experienced multidisciplinary team (BSPAR 2009; ARMA 2010). Emery (2004) considers rehabilitation to be vital and needs to be integrated into the comprehensive management of children with arthritis. Medications can control joint inflammation but rehabilitation needs to include maintaining range of movement. Emery (2004) further emphasizes the need to manage the condition from a holistic point of view and adhere to the programme assiduously. She notes that many young people have had their inflammation controlled after a period of time; however, they can be left with residual damage in a joint as a consequence of not adhering to the recommended physical therapy programme. There is a need to take a more innovative look at rheumatology service organization to ensure the smooth transition of young people from paediatric services to adult rheumatology services to reduce the attrition rates and promote good understanding of the rationale of the treatment regimen (Duffy, 2005; BSPAR 2009; ARMA 2010).

The overall objectives of care of this condition provide opportunities for multi-strategy health promotion initiatives in a variety of settings with special emphasis being placed on pain, coping strategies, physical and occupational therapy and nutrition.

Some young people with JIA may have been experiencing and coping with difficult challenges from an early age and consequently have exceeded normal psychosocial developmental ‘milestone’ expectations for their age (Shaw et al.,
2006; Östlie et al., 2009), while others may have been ‘overprotected’ and consequently there has been developmental delay. Some treatment regimen factors can impose lifestyle constrictions, particularly for young people in their late teens; for example, alcohol is contra-indicated if taking some medications.

### 2.9 Psychosocial Impact of Juvenile Idiopathic Arthritis

The aim of this section is to discuss the psychosocial impact of this chronic condition on the patient’s life, and consequently on their quality of life, and the impact on the patient’s family circle and beyond. Cognitive coping skills and the need for resources and being resourceful are discussed.

Juvenile idiopathic arthritis is a chronic disorder that may be self-limiting for some, but for the majority of young people with this condition there is currently no cure despite improvements and advances in pharmacological therapies and other interventions. The condition can pervade the patient’s life. Disease symptoms can disrupt (Dingwall, 1976), interrupt, intrude and cause uncertainty in all of the activities of daily living (Bury, 1982; Charmaz, 1991). The unpredictable nature of the disease in itself can be a stressor; for example, it can be difficult to make short-term social plans and if long-term plans are made, there needs to be an alternative plan to deal with the unexpected vagaries of the condition. Consequently, families who have a member with the condition need to be well organized and have an adaptable attitude to their circumstances. This is not always easy and the illness can place families at risk of psychosocial stressors. Stressors include access to health care, health insurance, quality of medical care, and socioeconomic status.

Traditionally the biomedical model focused on the care of the ‘sick child’. In recent years, however, the focus has been developed further to a biopsychosocial model to consider the ‘sick child’ within the family unit. Kiely et al., in a report prepared for the Commission on the Family (1996:13) suggest that ‘The experience of family living is the single greatest influence on an individual’s life’. The literature throughout the decades provided a number of
aspects of living in a family while living with the chronic disease of juvenile arthritis. Examples included learning to look after an ill child, adapting family life to meet the needs of the young person with JIA, and managing the treatment regimen. Commentators such as Strauss (1973), Strauss (1975), Jerret (1994), Britton (2004), and Shaw et al., (2006) have contributed a significant body of work in this area. In recent years there is a more person-centred and family orientated focus in the standards of care and in the provision of care (ARMA 2010; BSPAR 2009).

Within the family unit, the impact of a child having a chronic disorder can generate stress which may manifest itself as mood disorders in parents, overprotective parenting and the possibility of marital discord (Zimmermann, 1995; Noll et al., 2000; Barlow et al., 2002). However, Gerhardt et al., (2003) found families with a member with JIA to be resilient and not demonstrating signs of severe psychosocial distress.

Zimmermann (1995:301) suggests that ‘illness exerts a “centripetal pull” on the family system which increases in strength with the severity of illness and the degree of resulting incapacities in the child’. The coping strategies and adaptation styles of the family, as well as their available physical, practical and psychological support and resources, will influence strongly the ability and resilience of the family to cope not only with short-term crises but also with protracted periods of illness of their family member (Cavallo, 2009). Eiser (1994) suggests that these are ordinary families endeavouring to manage an extraordinary situation. Families with less adept coping strategies or who are more vulnerable due to their particular circumstances may be overwhelmed with endeavouring to manage a young person with JIA (Vuorimaa et al., 2009).

There are a myriad of cognitive coping strategies that can assist with positive adaptation. These include optimism (Scheier & Carver, 1985), humour (Brown & Hepple, 1989), an internal locus of control (Sloper & Turner, 1991) and help-seeking skills (Sloper & Turner, 1993). Research studies relating to parental relationships of a child with cancer highlight multiple sources of stress (da Silva,
2010) and the fluctuations of these stressors during the disease course. Stressors included anger, guilt and anxiety. Differences in parental coping strategies could give rise to parental upset and conflict. Some relationships were adversely affected by these stressors while others were strengthened (Yeh, 2004; Hentinen Kynas, 1998; Lavee & May-Dan, 2003).

Perrin, Ayoub & Willett (1993) consider ‘maternal mastery’ of the situation to be highly significant in promoting positive adaptive skills of not only their child with the chronic disorder but also other members of the family. Cuneo and Schiaffino (2002), in a study to assess the influence of disease activity, family resources and parental adjustment, reported that maternal self-perception of global self-worth was highly influential in providing positive adaptation to the situation. They also suggest that a positive and supportive family environment was likely to reduce the negative impact of the illness and greatly influence the young person’s psychological adaptation to the illness. This finding was consistent with previous research (Miller, 1999).

Britton (2002b) noted that the mother’s ability to inquire and retrieve information about services was directly related to the provision of services that the child received and not necessarily related to the severity of the child’s illness. Manuel (2001) notes that maternal education can be a significant factor in assisting mothers to advocate manage and cope with the stressors of having a child with a chronic illness. Vuorimaa (2009) suggests that research relating to psychosocial dysfunction amongst parents of children with juvenile idiopathic arthritis provides conflicting evidence. For example, Gerhardt (2003) suggests that there is no overwhelming evidence of it, while Barlow (2002) suggests that levels of anxiety/depression of mothers were high compared to a control group. Interestingly, there were few recent studies on the role of the father. Those available suggested his role was of a ‘silent’ supporter who tries to work harder to provide more resources for the family. This may reflect the role of some fathers with a child with a chronic illness. Providing extra resources for the family can positively impact on family coping strategies. For example, extra
financial resources can purchase specially adapted furniture to meet the young
person's needs, or it may purchase house-cleaning hours which could free up
the parents to spend more meaningful time with their children (Locker, 1994).
In general terms, however, the role of fathers has changed in this society from
previous eras, with many fathers now actively engaged in childcare and the
daily routines of family life (McKeown, 2001).

Britton and Moore (2002b) suggest that fathers have different priorities relating
to their child’s situation, for example the visibility of the condition and the
young person’s long-term future. Mothers prioritized the activities of daily
living and relationships with peers. Often fathers were not available during the
day due to work commitments to engage in care roles with the young person,
and when they came home children were frequently in bed (Timko 1992).
Staudacher (1991:3) suggests that men carry this loss of normality in their child
as a ‘solitary burden’. Despite family social arrangements having evolved in
more recent years, Waite-Jones (2008) suggests, that fathers concealed their
concern and their feelings even from their partner.

Research studies examining the relationship between problem-solving
strategies and adaptation report that families using problem-solving strategies
demonstrate less stress (Thompson et al., 1992; Kronenberger & Thompson
coping strategies and avoidance-coping or wishful thinking strategies, were
noted to have a greater number of stress-related issues.

Comprehensive assessment using a co-ordinated multidisciplinary approach
could assist with the provision of appropriate interventions to meet the
particular needs of the young person and their family with the objective of
assisting them cope with their circumstance and potentially reducing the
possibility of mal-adaptation (Zimmermann, 1995). The Arthritis and
Musculoskeletal Alliance (ARMA) (2010) refers to the significance of
empowerment and gaining the life skills and proactive behaviours to navigate life positively with chronic disease.

As a consequence of their condition, many young people with a chronic condition have potentially had more socialization with adults and in some respects their comprehension and reasoning may be more mature than their peers (Meijer et al., 2000). Consequently, it is important for healthcare providers and others to remind themselves on a regular basis of the need to treat each young person as an individual in a unique situation. This view correlates with that of Ungerer et al., (1988) who emphasize the need also, for using a developmental model for understanding the impact of and adaptation to living with a chronic illness.

Meijer et al., (2000), in a study to examine peer interaction in adolescents with a chronic illness, suggested that functional limitations were not associated with reduced peer interaction. Pain, however, was associated with reduced social interaction in boys. Meijer (2001) asserts also that neither pain nor functional limitations were risk factors for social adjustment problems.

Practical problems may be caused by rheumatic disease; for example, handwriting may be affected if young people have stiffness and pain in their hands. Chronic pain may also impact on sleep and cause fatigue, which may in turn lead to concentration difficulties (Schanberg, 2005). It is important that emphasis is placed on strategies to overcome practical difficulties to avoid educational difficulties that are also affecting quality of life.

Foster et al., (2003), evaluating quality of life in adults with arthritis from an early age, reported that many patients had active disease in adulthood. Foster also noted that physical outcomes were relatively good but despite good educational outcome many of the respondents were unemployed and this had had an adverse impact on their quality of life. This finding has been reported in other studies in the United Kingdom (Packham & Hall, 2002;) and in Canada.
(Kotaniemi et al., 2001). These findings have implications for healthcare providers and their strategies to promote health. Juvenile arthritis can affect a young person at many levels - physically, psychologically, emotionally and interpersonally.

When a child is diagnosed with a severe disease, the situation has the potential to distress the entire family network. Britton (2002) explains her three-phase model of families' experience of living with a child with arthritis. Building upon the work of Corbin and Strauss (1988), which explored the experiences of adult carers and chronically ill partners, it considered the illness trajectory. Britton also integrates the work of Gravelle (1997) and the concepts of normalisation and chronic sorrow (Olshansky, 1962; Anderson, 1981; Knafl and Deatrick, 1986; Worthington, 1989, 1994; Teel, 1991).

Britton's (2002c) three phase model:

**Phase one:** the family's experience is signified by anxiety, dealing with the symptoms of the disease, and seeking a correct diagnosis and treatment.

**Phase two:** the family is becoming more familiar with the pattern of the disease and endeavouring to manage the complex treatment regimen.

**Phase three:** the family and the young person become more experienced and more ‘expert’ in managing the situation and dealing with the predictable crises.

Britton explains that predictable crises within the model include, for example, the time of diagnosis and commencement of primary school or secondary school. These transition times can provide challenges for all families, but present particular difficulties when the young person has a chronic condition. Unpredictable times of adversity include periods of acute disease or other triggers related to the disease process. Britton also explains that each phase is unique to each family and will be impacted by multi-factorial variables,
including the family's internal and external resources, the disease process and the treatment regimen.

Shaw et al., (2006), discussing resources and their significance, makes the point that while physical resources and access to them is significant, being mentally resourceful, being creative and having problem-solving capacities are also extremely important.

Barlow et al., (1999) suggest that there is a need for greater consultation with the ‘experts’ in the context of juvenile arthritis; these are the young people with the condition and their parents. The experts indicate that they want more emphasis to be placed on the social and emotional aspects of chronic disease and the skills to manage the condition at home, at school or at work. Interventions could initially be focused on broad categories, for example mild, moderate and severe arthritis. Age and gender may also need to be categorized and specific interventions developed to meet specific needs.

Participation in decision making is also important and becomes more relevant as the young person becomes older. Historically ‘children were seen and not heard’. Today, children’s views are sought and clinical staff endeavour to facilitate children’s participation in decision making. This can have the effect of developing their own autonomy and also of influencing their own future and taking control of the management of their condition, and is aligned with the principles of health promotion (Runeson et al., 2002).

Care of the young person with JIA is quickly transferred to family members who frequently have concomitant demands on them, for example the care of other children or work. Parents, too, can be very anxious not only about the specific day-to-day care of their child but also about their child’s future well-being (Barlow & Ellard, 2006). Siblings’ lives can be disrupted, as outings, time with and attention from parents can be reduced (Shaw et al., 2006).
McAnarney et al., (1974) reported that teachers of children with more severe arthritis had fewer problems in making concessions for those more clearly severely affected children than children whose condition was apparently less severe. Children whose condition appears or is visibly less severe may have greater adjustment problems.

Family relationships are complex and so too are sibling relationships. However, Weiss et al., (2001) in a small-scale study (n=20) suggest that sibling relationships in which one child has juvenile arthritis is not a predictor of significant difference compared to the sibling relationship of a normative control group.

Noll et al., (2000), assessing the social, emotional and behavioural functioning of children with juvenile rheumatoid arthritis (n=74), found few differences between young people with juvenile arthritis and control children (n=74), indicating psychological resilience. This finding was unsupportive of disability/stress models of chronic illness in childhood. Noll et al., suggest the need for multicentre longitudinal investigations, but conclude that it is unlikely arthritis is the principal cause of significant emotional problems in young people with this chronic illness. Examining parent and child reporting of negative life events of young people diagnosed with cancer, juvenile rheumatoid arthritis, diabetes or cystic fibrosis Johnston et al., (2003) note significant differences between child reporting and parent reporting of the same life events. This has significance in many areas, not least in the provision of care for the young person and their family.

It is important to recognise that adapting to chronic illness is an evolving developmental process. The findings of Thon and Ullrich (2008) suggest there is a high level of interest of families seeking new and further information relating to the disease. The findings of Packman et al., (2005) and Békési et al., (2011) suggest the significant benefits of special summer camps for families of young people diagnosed with chronic diseases by providing opportunities for
family members to communicate their concerns in a safe environment. Greater appreciation of the condition may be gained, perhaps learning from other families also, thereby assisting families to adapt and cope with their particular situation.

Barlow and Ellard (2006), providing an overview of the current literature on the psychosocial impact of chronic disease on children, their parents and siblings, conducted electronic searches of the literature from 1990 to week 24, 2004. This resulted in six reviews of the psychosocial well-being of children being identified. Only one referred to juvenile idiopathic arthritis; two others related to asthma and sickle cell disease respectively and three were concerned with chronic disease in general. Four further reviews related to psychosocial well-being of parents (n=2) and sibling well-being (n=2). The meta-analyses demonstrated only a small risk to psychosocial well-being of the young people with chronic disease and their siblings. The evidence of risk to the psychosocial well-being of the parents is inconclusive. The authors concluded a more extensive evidence base was required to provide more informed information.

Waite–Jones and Madill (2008b:477) suggest that healthy siblings of a young person with JIA see their family as different to ‘normal’ families. They feel ‘inadequately informed’ about their sibling’s condition and that they ‘share vicariously adverse experiences of their ill sibling’. This exploratory study suggests the need for further research in this area of healthy siblings adapting to an ill sibling’s situation. The long-term sequelae of persistent disease into adulthood have many implications, including implications for healthcare providers (Zak, 2000).

Gutiérrez-Suárez et al., (2007), in the Pediatric Rheumatology International Trials Organisation (PRINTO) multinational quality of life cohort study, included 3,167 young people under the age of 18 in their study. A total of 30 countries were included. These were grouped into three geographic areas: 16 countries in Western Europe, 10 in Eastern Europe and four in Latin America. The study
Chapter 2: Literature Review

Acknowledged that socio-cultural differences can impact on perceived health-related quality of life; however, it concluded that compared with healthy peers, geographical location was not an issue. Young people with JIA in all geographic locations considered that their quality of life was affected by the disease and that disability and pain were the most significant stressors to impact on their health-related quality of life.

Many commentators discuss the impact of chronic illness on men, affecting not only their health status, their relationships with others but also impacting on their own sense of masculinity (Cameron & Bernardes, 1998; Schofield, 2000; Waite-Jones & Madill, 2008a). Healthcare providers need to assess the young person and their social functioning to identify vulnerable young people who may be at increased risk of internalizing problems (Sandstrom & Schanberg, 2004).

2.10 Healthcare Strategies for Young People with Chronic Diseases

Predicting and acknowledging the increase in the number of people to have chronic diseases, the World Health Organization has published a number of documents relating to the health care of young people with chronic diseases in which the challenges of chronic diseases are addressed (WHO, 2001; WHO 2005a; WHO 2007). The aim is to prevent the chronic diseases if possible by promoting healthy lifestyles through education strategies. If this is not possible the goal then is to reduce the risk of co-morbidities and support the young person in such a way as to enable them to enjoy and optimise their life opportunities. Another World Health Organization (WHO, 2005b) publication addressed the need to prepare a workforce to meet the challenges of provision of care in the 21st century. Current service organization, which is designed for a pattern of acute episodes of care, is seeing a shift to a greater need for provision of care for people with chronic illness. There is a need for health providers to develop and learn new competencies and expand and build upon their existing knowledge to meet a different pattern of care challenges.
The WHO (2005b) suggest that there is a need for more patient-centred care, and a greater need for partnership between the healthcare team and the patient to promote active participation, collaboration and co-management of the illness. Electronic media innovations are creating new and exciting modes of communication, including ‘tele-monitoring’ and ‘tele-health’ systems. Health policy responses to rising rates of chronic illness are endeavouring to address the needs of people with not only a single chronic illness but also co-morbid and multi-morbid illness (Aspin et al., 2010). Wagner et al., (2007) proposed a Chronic Care Model of care. Martin and Sturmberg (2009) proposed the complex adaptive chronic care framework. Others, for example West et al., (1997), proposed a model to meet the needs of a particular group of people.

The literature did provide strategies for transition of care from paediatric services to adult services for young people with arthritis; however, it did not demonstrate any models of care for young people with arthritis across the lifespan and across different settings.

2.11 Communications

The healthcare environment has changed considerably in recent decades. Many influences have created this evolving environment, including new and improved technology, patients’ expectations and consumer access to information. To communicate effectively with young people, care providers need to understand the learning processes and cognitive development of children and adolescents and have the appropriate communication skills.

The interpersonal relationship between healthcare personnel and their patients can be a complex one. It is a relationship of trust. It is also a relationship where the participants are of non-equal status, and where frequently issues of vital importance that are possibly emotionally laden are discussed and decisions made. Bensing (1991) refers to task-orientated or cure-oriented communication as belonging to the cognitive domain and care-oriented socio-emotional behaviour as belonging to the emotional domain. The doctor-patient
relationship is often involuntary in the case of children. Anderson and Zimmerman (1993), in a study to examine patient and physician perceptions of their relationship, report that when the relationship was viewed as a partnership, patients were more satisfied compared to physicians who had a more paternalistic approach to the relationship.

Initial contacts with medical personnel can be highly significant for adolescents. In a study by Britton (2001), 41% of all families indicated that the experiences they had had around the time of diagnosis strongly influenced how they had coped at a later stage. Parents indicated that conflicting advice from healthcare personnel and poor communication between them contributed to their distress. Britton suggests that healthcare personnel need to be more aware of the service needs of patients and families. Some studies have found that quality of life has been improved by patient education (van der Palen et al., 2001). Patient education has also been associated positively with compliance to health regimens and the development of positive coping strategies (Coates & Ryan, 1996; van der Palen et al., 2001). De Winter (2004) and Holtzheimer (1998) suggest that there is a direct correlation between compliance to the treatment regimen and the amount of time spent in direct doctor-patient communication.

These commentators suggest that this direct communication is positively associated with an improved understanding of the illness and of the patient as a person. Eiser (1990) noted that the deficit of knowledge amongst young people in relation to their chronic condition and its management was remarkable in its incompleteness and its lack of depth. Innovative ways of providing age-appropriate information and knowledge have implications for health promotion initiatives in this area. Kilkelly and Donnelly (2006:42) discuss the importance of engaging young people in participation in health education. They reported that the young people provided ‘principled and practical’ reasons for their views to be considered, including allaying fears and being prepared for situations or procedures. Being treated with respect and dignity, good humour, empathy and
kindness at all times were considered particularly important by young people (Sinclair, 2004; Reed et al., 2003).

Ullrich et al., (2002), in a study of German adolescents, reported that the males with JIA considered themselves less well informed about their condition than did their female counterparts with JIA. This finding has implications for clinical practice and health promotion, areas where there is already concern about the health of young men.

Passive involvement of young people in consultations with their physician is a regular occurrence. Davis (2006), acknowledging the difficulty of engaging a young child or young person in discussion about their condition, discusses the importance of developing a relationship from an early age with the young person in order that they participate in the consultations. Judd (2001), following a case study research project, suggests the need for healthcare providers to be watchful for signs of a mask of compliance, a mask of pleasant co-operation. Developing the young people’s communication skills from an early age will assist them to engage and contribute to healthcare team meetings enabling the young people to participate meaningfully in their healthcare provision decision making.

Clinicians need to be able to assess the young person’s cognitive development and be aware that cognitive development can be affected by many factors including age, loss of schooling during illness, impact of pain, fatigue, anxiety and depression. Concrete thinking and ‘here and now’ concrete explanations are needed for young people in early adolescence. Clinicians need to avoid more abstract communications, for example ‘if - when’ when talking to young people in early adolescence.

Barlow, Shaw and Harrison (1999), in a study entitled ‘Consulting the “experts”: children’s and parents’ perceptions of psycho-educational interventions in the context of juvenile chronic arthritis’, concluded that the social and emotional
aspects of chronic disease required greater emphasis. There was also a need for informal support groups as well as group information, which could go some way to reduce the sense of isolation and provide reassurance. Also, it was suggested that activity weekends and summer camps could be beneficial and would be welcomed. These finding were supported by Kyngäs (2003:751), who espouses the view that ‘each adolescent should be respected as an expert of her/his life’. Andre et al., (2001), in a study to evaluate changes in self-reported competencies following an education programme among parents of children with juvenile chronic arthritis (JCA) and among adolescents with JCA, concluded that information can assist in reducing parental anxiety and consequently reduce stress in the young people. This was particularly noted during medical procedures.

Britton et al., (2004) carried out a study to determine if adolescents with chronic illness want to make decisions about their treatment. Eighty-three adolescents with a mean age of 15.4 years and with sickle cell disease, cystic fibrosis, rheumatoid arthritis or inflammatory bowel disease were included in the study and completed a questionnaire to assess the adolescent’s priorities and preferences for health care. Contrary to the hypothesis of the research team, the majority of the adolescents preferred the treatment decisions to be made by physicians or that the decision making should be made equally. Participating in a decision making process has its own intrinsic value. It is recognised that there are different levels of participation (Hart, 1992) and frameworks to assess competency (Koocher, & Keith-Spiegel, 1990).

Shaw et al., (2006) carried out a study to examine the level of agreement between parents and adolescents with JIA. Areas to be compared included physical health, functional ability and health-related quality of life. The Childhood Health Assessment Questionnaire and the Juvenile Arthritis Quality of Life Questionnaire were used with visual analogue scales for pain and general well-being. Three hundred and three adolescent-parent groupings agreed to participate in the study. The study reported many differences in agreement
between the parents and the adolescents with JIA. Acknowledging that perspectives differ, the study has implications for clinical practice, not least in the area of communications.

Young people seek a health service which views “adolescents as people rather than problems, and (is) able to tailor practices to individual adolescents” (WHO, 1999:95). Beresford and Sloper (2003), in a qualitative study to explore chronically ill adolescents’ experiences of communicating with doctors, found the young people wanted choice as to who was present at the consultation. The gender of the doctor was also significant; a sense of seeing the young person as an individual and not just a condition was important. Continuity of contact with the same physician was also considered to be significant. The young people also noted that the presence of a parent could constrain the development of a doctor-patient relationship.

Reed et al., (2003), discussing the concept of dignity of the child in hospital, emphasized the significance of promoting this concept in all settings. Reed et al., emphasized the need of promoting the dignity of the child both the macro dignity and the micro dignity of the young person. The intrinsic worth of the person and their best interests (Kopelman, 1997), including their personal privacy, need to be maintained as priorities by carers (Beauchamp & Childress, 2001).

Mayall (1996) discusses how children are required to comply with adult control. Consequently, power issues can be a stressor in a family. Lansdown (1994) makes that point that children’s experiences in relation to their bodies and their lives are very different to those of adults. These issues are very pertinent in the healthcare situation. It is, however, important to make the point that this very vulnerable population have a high standing in legal terms (Mills, 2007), including the European Convention on the Exercise of Children’s Rights (Council of Europe, 1996) and the Convention for the Protection of Human Rights and
Dignity of the Human Being with Regard to the Application of Biology and Medicine (Council of Europe, 1997).

There is a need to assist patients and families to understand the disease process; what is happening to their body, how it is happening and what is the best course of action to control it and avoid damage to joints. Perhaps more user-friendly anatomy and physiology information should be available on the 'mechanics' as to why joints assume 'positions of comfort', which are usually flexed positions leading to muscle spasm and shortening, with consequent contractures and 'the domino' impact on other muscles, which try to accommodate and compensate, leading to increased energy expenditure because muscles are working within a compromised range only.

If young people and their families fully understood 'the mechanics' of joints and the implications of less than optimal adherence to physiotherapy programmes, there may be less difficulty with compliance to these programmes. Interestingly, there appears to be a paucity of information on this significant issue in the literature.

In conclusion, communication skills need to be age- and developmental stage-appropriate. The skills used with teenagers are different to those used with adults. Education and training for personnel working with teenagers in clinical situations needs to be analyzed and reviewed.

2.12 Social Support
The aim of this section is to discuss the significance of social support.

Social support can be defined as the 'processes by which interpersonal relationships promote well-being and protect people from health declines, particularly when they are facing stressful life circumstances' (Lanza & Revenson, 1993:97).
Many authors associate social support with positive well-being for adolescents with a chronic disease (La Greca et al., 1995; Price, 1996; Kyngäs, 2000; Kyngäs 2004). The key members of the social network include family and friends. Good compliance with a treatment regimen is correlated with positive family dynamics, while discordant family relationships are associated with poor compliance (La Greca et al., 1995; Miller-Johnson et al., 1994; Kyngäs, 2000).

Berntson, et al., (2003) comment on the heterogenicity and the severity of juvenile arthritis. Juvenile idiopathic arthritis is a self-limiting disease for some patients but also a debilitating progressive condition for others. They assert the importance of identifying early those with the more severe forms of JIA in order to be 'situation aware' in an effort to reduce the impact of potential problems physically and psychosocially. They emphasize also the need to provide the necessary support for those with milder forms of JIA. Britton (2001) indicated that families may need support due to the oscillating nature and unpredictability of the condition contributing to family distress and consequently impacting on family quality of life.

Friends and peers can provide emotional support through acceptance of the young person and by their inclusion in the peer group. This can be associated with better coping skills and may have a buffering effect, but the opposite can also be true. Vilhjalmsson (2003) refers to the ‘strain hypothesis’: as a consequence of insufficient social support the impact of the disease and distress may be increased. This is also known as the vulnerability hypothesis. Kyngäs et al., (1998) advises that frequently peers have a positive impact and often can assist the young person with a chronic condition. However, there are times when peers can exert a negative influence; the young person with the chronic condition may want to have the same lifestyle as their friends and concordance with a recommended treatment plan is abandoned.

In a qualitative study in Finland, 40 adolescents with a chronic disease were interviewed. The findings indicated that not only were family, friends, peers,
healthcare personnel, school and other adolescents with a chronic condition important for providing social support because each had a unique role, but also important to the support network of the young person were pets and technology. The young people considered that their pets understood their emotions, had a constant presence in the home, and were available when the young person needed them. They also reacted appropriately (Kyngäs, 2004).

Noll et al., (2000), in a study to investigate if young people with arthritis have more social and emotional problems than case-control classmates, found a psychological resilience amongst young people with arthritis. It was also noted that the fact that the young person had arthritis was not the principal factor in situations where significant social or emotional problems were evident.

Packham and Hall (2002) carried out a study to assess the level of education attained and employment gained in a group of 246 adults identified with an average disease duration of JIA of 28.3 years. A comparative group of their siblings and national statistics was used. The findings were very interesting. The study group achieved significantly better results than the national average and their siblings. The unemployment rate, however, in the patient group was twice that of the national average. The incidence of workplace discrimination was found to be 25:1. Packham and Hall (2002) note the importance of social attitudes towards disability. In recent years Ireland was the host nation to the Special Olympics and there is no doubt that the event raised disability awareness at the time. Recent legislation (The Disability Act 2006) has also increased disability awareness.

Packham and Hall’s study (2002) also highlights the significance of transition from education to employment and the importance of educational achievement and coping skills. The need for specialist career guidance was evident; interview techniques and assertiveness training were recommended. Discrimination by omission and commission were found by the patient group, problems at job interviews (45%), and once in a job overt and covert
discrimination was encountered, with 26% being passed over for promotion or sidelined into less fulfilling work. Financial security and independence rely heavily on being employed. Psychological health too can be significantly affected by unemployment.

Ongoing accessibility to expert advice can be of enormous value, particularly during a flare-up when direct access to expertise can positively impact on reducing the intensity of the flare-up and accelerating recovery. Knowing that the situation is being monitored even at a distance, can provide relief and comfort to families. New technology and greater use of multimedia electronic communication can assist with this form of service provision and service development.

Residential workshops for parents of adolescents with juvenile idiopathic arthritis can provide a safe environment for parents to discuss their concerns and have been shown to be an effective intervention in the care of young people with JIA (Turner, et al., 2001).
2.13 Transition to Adult Services

Adolescence is a time and a process of transition from childhood to adulthood when young people develop their life skills and competencies and assert their independence by establishing relationships outside the family, finding employment, and forming a personal and sexual identity (Packham, 2004). It is a time of change and challenges; however, some adolescents with a chronic illness may not have the same opportunities or abilities as their healthy peers.

The transition from childhood to adulthood is a process that requires a number of milestones to be met (Hardoff & Chigier, 1991), including the consolidation of personal and sexual identity, the establishment of relationships outside the family, the achievement of independence from the family, and the finding of a career/vocation.

In recent years there has been greater recognition of the particular needs of adolescents with chronic conditions and healthcare provision. Transitional care is defined as ‘a multi-faceted, active process that attends to the medical, psychosocial, and educational/vocational needs of adolescents as they move from child- to adult-centred care’ (Blum et al., 1993:570). The process of planning transitional care requires a collaborative effort from the young person and the healthcare team. Transitional care is advocated by many commentators (BSPAR 2009; ARMA 2010). Blum et al. (1993) suggest the need for a continuum of care from the paediatric care team service to the adult care team service. Paone et al., (2006) have integrated a multidisciplinary approach to their model of transitional care, the ON TRAC (Taking Responsibility for Adolescent/Adult Care) model, which advocates early planning and the provision of sustained opportunities to develop life skills.

Rosen (2003:309), in a position paper of the Society for Adolescent Medicine on transition to adult health care for adolescents and young adults with chronic conditions, states that the goals of a co-ordinated transfer to adult health care are:
To optimize health and to facilitate each young person's attaining his or her maximum potential.

Proactive transition to the adult healthcare system encourages young people to be successfully integrated into a comprehensive care system to meet their complex needs.

The transition must include primary, secondary and tertiary care with a careful delineation of a financing system that will support a comprehensive care programme.

Rosen (2003:309) suggests that the principles of successful transition include the implementation of age- and developmental stage-appropriate services. Care should meet the unique needs of the individual. Also, a transitional programme should include services that would concern all adolescents, including substance abuse and other health promoting and damaging behaviours.

Rosen makes the point that transitional care is most successful when a co-ordinator who will liaise with the patient and the family and the healthcare team is appointed.

Recognising the possibility that adolescents with chronic conditions may have psychosocial delay and may display more dependency behaviours, transitional programmes may enhance self-reliance, personal responsibility and autonomy.

Packham et al., (2002) noted that patients developing JIA over the age of 12 years had the highest risk of developing anxiety-related problems.

Paone, et al., (2006) discuss the importance of successful transition from paediatric services to adult services. Developing the competencies and life skills to actively engage and participate in their own healthcare management requires preparation. This is a process that needs to be developed at an early age. It needs to be co-ordinated, developmentally appropriate, psychologically sound
and uninterrupted. The ON TRAC model (Taking Responsibility for Adolescent/Adult Care) is a generic model of transition care for adolescents with chronic diseases and provides a clinical pathway. There is a need for a coordinator to liaise with the young person and the care team. Documentation should include a portable medical record, a transition record and the healthcare plan for the future. Transitional care services need to be more than transferring the patient and their records to adult services. Pai and Ostendorf (2011) reviewed the literature relating to the transition process from paediatric services to adult services amongst young people with chronic diseases. The rates of attrition and factors that impact on the process were analyzed. They highlighted that high rates of non-adherence to medical regimens (Annunziato, 2007; Kipps et al., 2002; Martinez et al., 2000) that can occur at these times result in difficulties with physical (Kennard et al., 2004), psychological (Drotar, 2007) and quality of life issues (Ekberg et al., 2007; Fredericks et al., 2008).

Barlow et al., (1998) reviewed seven studies relating to psychosocial interventions for children with JIA. None of the studies were RCTs and five had samples of less than 13 children. Within these limitations cognitive-behavioural therapy, summer camps and family retreats were all found to be beneficial. Mentoring was also found to be of assistance to mothers of children with JIA. The mentors were mothers of young adults diagnosed with JIA during early childhood. The mentoring intervention evaluated by Ireys et al., (1996) demonstrated an improvement in maternal mental health and social support compared to the control group.

Active self-management is associated with higher adherence to treatment regimens, improved outcomes and high levels of self-efficacy. However, chronic diseases are frequently unpredictable. Fluctuations and crises are part of chronic diseases and can take their toll, causing people to have higher levels of fatigue and pain. These can have implications for the person’s ability to socialize and to take exercise. The process of recovering from a flare-up of the
disease can take a number of months during which the young person may have reduced energy levels and stamina. These can impact on the quality of a person’s ability to interact with others, socialize and work at school with a potential for isolation, leading to low mood and possible depression, appropriate support is needed to minimize the sequelae of a ‘flare-up’ (Shaw 2006).
3. ETHICAL CONSIDERATIONS

The aim of this section is to discuss some of the ethical considerations that guided the project. Principles within the Nuremberg Code (1947), the Helsinki Declaration (World Medical Association, 2008), and the Data Protection Acts 1988 and 2003 are discussed. Consent, including informed consent, ongoing consent, voluntary consent and the capacity to consent, is a key issue. The importance of children’s views and concerns is considered and addressed. The key element of the ethical guidelines referred to the dignity of the person, respect for the person and safeguarding the best interests of those participating in the research study.

The Belmont Report (The National Commission for the Protection of Human Subjects of Biomedical and Behavioural Research, 1978) provided research ethical principles to help safeguard research participants and assist researchers. Beauchamp and Childress (1979) in their seminal work discuss the importance of justice, autonomy, beneficence and non-maleficence. The principle of justice is a broad concept and is associated with fairness, equity, honesty and correctness (for example, has the person made the right decision?). The principle of autonomy is concerned with the obligation to respect the person as an independent individual with the right to self-determination (Rogero-Anaya, 1994). Beneficence is associated with the positive potential of doing good, for example the benefits of taking part in the research. Mulhall (2001:541) poses a number of questions relating to the therapeutic versus the research imperative. ‘Is it deontological, where the individual is not a means to an end but an end as such? Is it advocacy for human rights? Is it based on justice, beneficence and respect for patients’ rights? Is it utilitarian, where people are used as a means to further knowledge?’ These are some of the issues that are posed and require exploration and analysis.

Non-maleficence refers to the obligation of the researcher to avoid harm. The principle of non-maleficence resonates with the Hippocratic oath of ‘do no
harm’ (Swinton, 2009). Other issues of concern include the fact that the benefits of taking part in the research must outweigh the risks. The aims of the research, the methodological approach of the research, and the outcomes of the research need to be considered from an ethical standpoint for robustness in relation to the rationale of the project, and as to how the project will increase the body of knowledge on the topic (Burns & Grove, 2005). Veracity is about the honesty of the work, and the responsibility of the researcher to provide honest information and explanations for participants and honest interpretations of the data. Fidelity is associated with trust, and its significance in research relates to the necessity of researchers to protect participants from risk (An Bord Altanais, 2007). Many commentators discuss the significance of ensuring that the ethical aspects of a research project and its potential implications are considered in depth before the research is conducted (Field and Morse 1996; Ross et al., 2010).

Practical, physical and psychosocial implications for participants need to be considered. Exploration of potential unforeseen, unintended consequences of the research and the contingency strategies to manage such situations also need to be considered prior to data collection. Hammick’s (1996) ‘research ethics wheel’ outlines research ethics issues for consideration and provides a useful practical guide. Cutcliffe and Ramcharan (2001) discuss the need for an ‘ethics in process’ model, suggesting the need for an ongoing consent process, checking periodically with participants if they wish to continue to participate. The research process is a dynamic process, and consequently a participant may have consented to participate in the study within particular parameters or circumstances. If, however, those parameters or circumstances change or are revised the research participant needs to be advised in order that they too can revise their decision to consent to participate in the study.

The International Council for Nursing (ICN) (1996) also provides a framework of ethical principles for nurses carrying out research. These include beneficence, non-maleficence, fidelity, justice, veracity and confidentiality.
These principles have been further refined to four areas, which are: the right not to be harmed, the right of full disclosure, the right of self-determination (subject’s right to decide to take part or to withdraw at any time), and the right of privacy, anonymity and confidentiality (Howarth & Kneafsey 2007).

3.1 Children as Participants in Research

The United Nations Convention on the Rights of the Child (United Nations, 1989) is clear in its indication that children need to be involved in decisions that impact on them. This has been reinforced in legislation in other countries, for example the Children’s Act 1989 (England) and The Children (Scotland) Act 1985. Many bodies provide ethical and practical guidelines for the use of researchers, including the World Medical Association’s Declaration of Helsinki, which was amended at the 59th WMA General Assembly (2008) and provides ethical principles for medical research. More specific guidelines for children include ethics guides from the Medical Research Council (2004), the National Children’s Bureau UK (2003) and Barnardos (2002). The National Research Ethics Service UK (2007) provides information sheets and guidance for researchers and reviewers (Twycross, 2009). In recent years in Ireland, there has been greater recognition of the needs and the rights of children (Considine & Dukelow, 2009). There have been many changes in child care policy within a relatively short timeframe of twenty years approximately. A social policy framework for children began to emerge with the implementation of The Child Care Act in 1991, and in 1992 the ratification of the UN Convention on the Rights of the Child by Ireland occurred. Other developments in social policy in recent years demonstrate greater appreciation of the needs of children to be protected by government policy. In 1994 a dedicated Minister of State for Children was appointed. This led to the publication of the first National Children’s Strategy in 2000, having consulted with 2,500 children and young people. Following on from this publication and growing recognition of the need for child protection the National Children’s Office was set up in 2001 to implement the National Children’s Strategy (2000) and the office of the Ombudsman for Children was established in 2004.
In Ireland, The National Children’s Strategy (2000:30) recognises the need to provide children with the opportunity to provide their opinions on areas of interest to them. One goal in this strategy document states: ‘Children will have a voice in matters which affect them and their views will be given due weight in accordance with their age and maturity’. This statement indicates the relevance of consulting with children and the importance of listening to their views and concerns.

The Department of Health and Children (2009) published a document relating to children’s research, *Children’s Research & Ethical Review*, which recognises the significance of children’s research. It indicates the need for a culture of safety and protection for children and the need to have a national governance body with the authority to implement national standards for research with children. There is acknowledgement of the need to protect children as they are a vulnerable section of the population. However, it is recognised that there is a need to improve practice and care as a consequence of consultation with children. The rationale for participation of children in a research study must be to increase knowledge and improve practice so that children can benefit from subsequent developments in health care (McGuinness & Wilkinson, 2009).

Many of the guidelines refer to the need for security screening of researchers prior to commencing the research (MRC, 2004; NCB, 2003). The limits of experience and expertise of the researcher should be acknowledged (Barnardos, 2002; NCB, 2003; RCN 2009). Preventing and avoiding harm is advocated. The National Children’s Bureau (2003) suggests the need for a period of debriefing prior to the end of an interview meeting in an effort to identify any adverse effects of an interview. The exchange of contact details is also recommended in an effort to provide sources of help or assistance for the young person post-participation in a research study. One example of safeguarding all of the participants in the study, particularly the younger participants under the age of 18 and the researcher, is to detail the venue of the interviews and those present during the interviews.
3.2 Informed Consent

The aim of informed consent in research is to allow potential research participants make informed choices as to whether they wish to participate in the research. Participants need to be fully aware of the process of the research study and the decision to participate must be voluntary. The Nuremberg Code describes informed consent (1947) as follows:

'The voluntary consent of the human subject is absolutely essential. This means that the person involved should have legal capacity to give consent; should be so situated as to be able to exercise free power of choice, without the intervention of any element of force, fraud, deceit, duress, over-reaching or other ulterior form of constraint or coercion; and should have sufficient knowledge and comprehension of the elements of the subject matter involved as to enable him to make an understanding and enlightened decision.' (Boomgaarden, 2003:108).

The British Educational Research Association (BERA, 2004:6) defined informed consent as ‘the condition in which participants understand and agree to their participation without any duress, prior to the research getting underway’. Consent, to be real or valid, must be voluntary, informed and given without undue influence or duress (Van Dokkum, 2011; BERA 2004). Didcock (2006) discusses the importance of informed consent and its particular significance when related to young people, who are considered to be a vulnerable population. Informed consent in this project meant that parents of the child gave written consent following appropriate information giving. Young people under the age of 18 in Ireland may not have the legal capacity to give informed consent; however, they may give informed assent (Mills 2007; Kirk, 2007). The children agreed to give their written informed assent agreeing to take part in the research following information being provided in an age- and cognitive development-appropriate format. Participants needed to have the capacity and the competency to understand and disseminate the information provided in order that they could make an informed decision. The group were vulnerable,
and the possibility of coercion or a sense of obligation to comply with requests to participate in the research project also needed to be considered (Hinds et al., 2007; Alderson & Morrow 2004). Hinds et al., (2007) make the point also that caring, sympathetic behaviour of the study team members may make refusal more difficult. All of these issues required careful consideration. Endeavouring to get the study completed there was the tension between being assured that all participants were properly informed and were giving their voluntary consent/assent, and meeting the young people and hoping that they were not going to be affected negatively by participating in the study.

3.3 Anonymity and Confidentiality
Anonymity and confidentiality are assured under normal circumstances. However, in legal terms the guarantee is not absolute and may be overridden, but only in very particular circumstances of potential or real harm to the young person or others (Van Dokkum, 2011). The best interests of the child must be of primary importance always. The researcher must disclose the information to the appropriate authorities if the child or others are at serious risk of harm.

3.4 Gaining Ethical Approval for the Study
To commence working on the project ethical approval needed to be sought. Each hospital site required individual approval from their own local research ethics committee to proceed with the study. Gaining ethical approval from each hospital site for the project required submission of a detailed written application. This application included the named support of a local medical consultant who would be the named principal investigator on site. Signed approval from a university academic supervisor was also required. All information to be given to the respondents, including letters of introduction, leaflets, consent forms and questionnaires, were required to be submitted as part of the application. Ethical committees reviewing research applications are voluntary and may only meet on a bimonthly basis, consequently, time delays can occur. This situation arose in relation to this study on three sites. As the
applications were not standardised the requirements for each committee were slightly different.

One site required Garda clearance. This was one of the first sites to be applied to for research ethical approval, and consequently on subsequent applications, it was useful to be able to state that Garda clearance had been gained (please see Appendix 5). In recent years Garda clearance is being sought increasingly for persons working in many settings, but particularly for those working with children. Gaining Garda clearance can be a lengthy process due to increased applications and level of activity in this office. This situation is a significant factor in the timeline of the data collection period.

Two research ethics committees also sought a personal interview, which included a presentation providing an overview of the project. Detailed questions were asked about the project, including the potential for harm and the strategies in place if it emerged that a young person had become upset by some of the questions or if it emerged in the questionnaire data that a young person(s) was adversely psychologically affected by their arthritis condition.

All of the applications for ethical approval of the project sought details as to how these ethical principles were being addressed in the project, including issues relating to compliance with data protection legislation. For example, questions were asked relating to where the data were to be stored and how the data were stored. It is noteworthy that all of the consultant physicians (the principal investigators) checked with me that ethical approval had been gained prior to commencing data collection at all of the sites (please see Appendices 6a – 6h).

The consent form needed to comply with the university requirements and also local requirements. Consequently, minor amendments were made to meet the local requirements of different sites. Consent is only valid if it is informed and voluntary. It is a process where the details of the research, the purpose, the process, the intended outcomes, the potential risks and benefits and all the
relevant issues are disclosed to the potential participant. It is important that people do not feel obligated to participate. The researcher, when seeking participants, needs to emphasise that participation is voluntary, particularly with a vulnerable population of young people under the age of 18 (Parahoo, 1997). Consideration needed to be given not only to the parents of the young people with arthritis but also the young people themselves. This was done using letters of introduction and information relating to the project, and also talking to and consulting with the young people and their parents. The presentation and the use of language of the information leaflets were developed to meet the needs of the young people as well as adults in an age- and cognitive development-appropriate format. The nature of consent is that it is informed, voluntary and ongoing and the person has the capacity to make decisions. The participant may wish to withdraw from participation if the information or the circumstances change. It was important to check with the young people that they understood what was being requested of them and that they were willing to participate in the research, whether this was taking part in an interview or completing a questionnaire (Masson, 2004). The age of legal capacity is normally 18, with some exceptions. If under the age of 18 the young people may not be able to give written legal consent but they can provide written informed assent. Parents of participating young people were asked to give their consent and the young person provided their assent in writing. All of the young people who were approached were very interested in the project and provided their written assent.

Data collected needed to comply with the Data Protection Acts of 1988 and 2003. Data were anonymized and contact details of participants were kept separately.

3.5 Summary
Ethical issues were considered to establish implications for the study. The project was guided by the ethical principles of autonomy, justice, beneficence and non-maleficence. The safety and well-being of all concerned in the study
were of critical importance. Careful ethical consideration was required to identify concerns before commencement of data collection. Consent from participants needed to be voluntary, informed and ongoing and the research participants needed to have the capacity to consent. The relevance of the research, pertinent information for potential participants and the nature of participation were significant issues for informed decision making by participants. The children participating in the research added another ethical dimension and further reinforcement of the need to safeguard the well-being and interests of all those involved in the research. The importance of respecting the rights and dignity of participants and the time and effort contributed to the research study by the participants needed to be acknowledged as a dimension of the ethical framework. The role of research ethics committees is to safeguard and protect the health, welfare and rights of participants in research studies. Each site had local compliance requirements to minimize predictable risk to participants.
4. DESIGN APPROACHES METHODOLOGY

4.1 Introduction
This exploratory quality of life study used both qualitative and quantitative methods. Data source triangulation and method triangulation were used in the study. It was applied research and it was underpinned by a pragmatist paradigm. The aim of this chapter is to provide a context and an understanding of the theoretical framework that underpinned the study. The chapter considers the rationale for choosing particular concepts to provide a foundation for the study, the ideas and concepts that initiated them, how they were framed and how they were developed.

4.2 Historical Perspective
Historically, different types of research have distinct principles and distinct world views. Mixing different types of world views in a research study can be contentious; therefore, the rationale for using mixed methods needs to be strong and needs to be well considered to avoid undermining the study. Some of the aspects of quantitative and qualitative methodology are examined to acknowledge the different stances of each and to assist with the rationale for a mixed methods approach to the study.

The terms ‘quantitative’ and ‘qualitative’ are used to describe two world views or paradigms in research. These are linked to their own separate ontological, epistemological and axiological underpinning assumptions and their own distinct methodologies, including their strengths and weaknesses (Willis, 2007). Quantitative research gives rise to a formal systematic, logical positivist/scientific world view. Qualitative research is linked to an interpretivist/constructionist, naturalistic, humanistic view with a post-positivism foundation.

The intent of quantitative research is objectivity, detachment and control. The process aims to reduce the sum to its parts and explain the parts, sometimes
referred to as a ‘hard science’. In quantitative research the theoretical focus is on testing theory. Knowledge is generated by deductive reasoning, which ‘moves from the general to the particular’ (Fieldman, 1998:137). Objective and quantitative data are concerned with measurement and establishing the relationship between selected factors. Subjective and qualitative data are concerned with understanding people's experiences of the world (Cohen & Manion, 1989).

The focus of qualitative research is to provide holistic meaning and understanding on a macro- and micro-level and emphasise the role of language (Dodd, 2008). The theoretical focus is on developing theory. Often the design in qualitative research is flexible and emergent (Frankel & Devers, 2000). The reasoning in qualitative research is dialectic and inductive. Inductive reasoning moves ‘from the particular to the general’ (Fieldman, 1998:137). The researcher shares in the interpretation of qualitative research; this, too, has an impact on the research. Guba and Lincoln (2005) refer to this type of methodology as hermeneutical. Qualitative research can be regarded as more subjective and is considered by some authors as a ‘soft science’ (Burns and Grove, 2011).

4.3 Rationale for the Utilization of Mixed Methods Methodology

Some commentators believe that research studies should be either quantitative or qualitative and that mixing them is incompatible with the core origins of the epistemology, paradigms and frameworks from which they were derived (Rossman & Wilson, 1985; Howe, 1988). However, other commentators suggest that ‘rather than thinking of qualitative and quantitative strategies as incompatible, they should be seen as complementary’ (Malterud, 2001:483). Teddlie and Tashakkori (2003) suggest that greater understanding of a phenomenon can be facilitated by utilizing a number of perspectives to study it. Baum (1995) suggests that an array of instruments is required to explore complex issues. This can include both qualitative and quantitative methods. Morse (2003) suggests that if a mixed methods methodology is used each
method should adhere to its own distinct guiding principles to benefit from the strengths of the method. Issues raised and questions asked related to the appropriateness of mixed methods methodology, in other words would it answer the research question? Would it add value to the research or would it weaken its strength? Patton (1990) is of the view that there is a need to understand the reality of life in all its complexity. The research question is of primary importance and utilizing the appropriate methods to answer the question is vital. The researcher should acknowledge the distinctions between the two schools of thought but should forego the quantitative versus the qualitative debate, move beyond it and adopt an impartial approach to it. Patton’s views brought the methodological discussion back to the project at hand and re-focused it on the aim and objectives of the study and the issue of the most appropriate approach to the study.

### 4.4 Methods That ‘Fit’ the Research Question

The aim of the project was to endeavour to gain a deeper understanding of the quality of life of young people with arthritis by evaluating their perceived health-related quality of life. Embedded into the ongoing planning of the project were considerations not only of world views but also the appropriateness of the methods in the research process that were to be used with young people and adults, to avoid harm and to endeavour to accurately reflect the views of those taking part in the research project. Quality of life analysis has its roots in social research assessing living standards. The purpose of using quality of life analysis is based on practical applications; by improving knowledge in the area it can influence policy issues.

The domains of assessing quality of life are constructed in consultation with potential respondents when generating instruments for use. Sarantakos (2005:326) suggests that quality of life analysis is frequently studied within a quantitative research model ‘leading to results that are not only accurate and precise but also generalisable and comparable’. Qualitative evaluation can also be used through the use of interviews and personal reflection but the results
offer ‘limited generalisability’. A pragmatic approach could use both qualitative and quantitative methods if they were the most appropriate methods for the phenomena under study (Onwuegbuzie & Leech, 2005a).

### 4.5 The Appropriateness of Pragmatism

External issues influenced the theoretical framework of the study also. The need for flexibility, adaptability and pragmatism, it could be suggested, were ‘thrust upon’ the study. The intention of the study was to explore the quality of life of young people with arthritis. In the early stages of the project it was envisioned that the study would be a predominantly quantitative study with only a small amount of qualitative work in the preliminary phase to assist with the generation of questions for a health services questionnaire. However, as the project evolved it unfolded that the expected numbers of young people with juvenile arthritis to complete the questionnaire could not be sourced.

The design of the study needed to be reviewed and revised and a new, adapted approach taken. After due consideration, a pragmatic approach emerged. This approach would, however, change the research design from a quantitative dominant design to a qualitative dominant design. A decision was made to interview a number of parents of children with JIA on the western seaboard of Ireland and to interview members of the clinical team in the same area to gain their views on the quality of life of young people with juvenile arthritis. It is important to acknowledge how the conceptual framework and the world view can be and was impacted to meet a ‘real world’ situation. If the study was to move forward, problem-solving skills and a flexible and pragmatic approach to its design were required.

Collecting data from a number of sources and by more than one method allowed the construct of health-related quality of life of young people with arthritis to be viewed from a number of perspectives - young people, their parents and clinicians. The aim was to increase the richness of the data by the use of triangulation. Knafl and Breitmayer (1989:210) explain that triangulation is ‘a
technical term used in surveying and navigation to describe a technique whereby two known or visible points are used to plot the location of a third point. The term was found in the social science literature as a metaphor in the 1950s to explain the use of multiple methods to measure a single construct. In more recent times the term refers to ‘the research practice of combining methods within a single tradition (quantitative or qualitative) or across those traditions’ (Boyd, 2001:580). Denzin (1978) indicates that there are four types of triangulation: theoretical, data, investigator and methodological triangulation. Data and method triangulation were used in this study.

Some of the conceptual paradigms of phenomenology resonated as being pertinent to the study also. Benner (1985:1) considers quality of life within a phenomenological perspective and comments on the need to capture the unique contextual, experiential perspective of the research participant. Mills (1994:27) states phenomenologists seek understanding of an experience, not understanding of the concept, and the overarching question is ‘what is the meaning of one’s lived experience?’

The concept of verstehen (understanding) is concerned with understanding the subjective perceptions, opinions and realities of daily life of participants of the research. It is concerned with what has guided this process of how and why their reality is perceived (Sarantakos 2005). The concept of verstehen is context-sensitive. It is interested in how people make sense of and interpret their particular situation and why people have constructed their views and opinions and interpreted their reality, for example a teenager with arthritis having the social support of good friends. Palmer (1969) suggests that interpretation is bringing understanding to the process. It is acknowledged that this study has been influenced by phenomenological conceptual thinking.

This literature was useful to explore the depth and breadth of quality of life and how it is interpreted within its context.
4.6 **The Rationale for Purposeful Sampling**

Purposeful sampling was used in the study to provide information-rich sources of data. The sources of information included the young people with juvenile arthritis, their parents and their clinicians. Patton (1990:160) explains the significance of purposeful sampling. ‘The logic and power of purposeful sampling lies in selecting information-rich cases for study in depth. Information-rich cases are those from which one can learn a great deal about issues of central importance to the purpose of the research, thus the term purposeful sampling’. The population of young people with arthritis is small. The population to meet the study criteria is smaller.

Rare populations are often accessed by ‘snowball’ sampling or by convenience sampling. Neither of these methods would have been possible due to gate-keeping mechanisms in place. Speziale and Carpenter (2003), discussing purposeful sampling, emphasise the need for emerging concepts to be represented. This requires research participants who are central to and have information relating to the phenomena being studied. Chang et al., (2009:839) suggest ‘that numbers do not provide the “power” in purposeful sampling the way they do in probability sampling’. Chang et al., are of the view that it is more useful to explain the engagement and interaction with participants of the study, such as participants’ observations to support an interpretation.

The validity of the study was dependent on participants with rich sources of information. However, it could be considered that this created a situation whereby the sample was ‘deliberately biased’ instead of accessing ‘interesting possibilities’ (Wood & Christy, 1999:189). The two key reasons for using purposeful sampling in the study were the exploratory nature of the study, which required access to sources of rich data amidst the challenges of accessing a small population, and the need for pragmatic decision making in the design of the study.
4.7 Summary of Theoretical Framework

The exploratory nature of the research question shaped the theoretical framework, methods and sampling of this quality of life study. The design needed to be fit for purpose. Issues that needed to be addressed included the appropriateness of the methodology to answer the research question and if the methodology would add value to the study or weaken it. Other issues related to the results and the possibility of them being constrained by the use of one method only.

Triangulating the data and the methods allowed for the potential of complementary data to enhance the understanding of the phenomena under study. It also served the purpose of improving the validity of the study. The ‘real-world’ context and external issues to the study further promoted a pragmatic approach to the theoretical framework methods in social and behavioural research.

Aims and Objectives of the Study

The aim of the study was to explore the health-related quality of life of young people in Ireland with juvenile idiopathic arthritis (JIA).

Objectives:

- To explore the health-related quality of life of young people with JIA and compare their views with a peer group.
- To explore the health services experiences of young people with JIA.
- To explore the perceptions of quality of life issues of parents of young people with juvenile idiopathic arthritis
- To explore the perspectives of clinicians in respect of quality of life issues for young people with JIA.
4.8 PHASE 1: Consultant Survey

Phase 1 Introduction
In the absence of a juvenile arthritis registry, the aim of phase 1 of the project was to estimate the number of young people with arthritis in Ireland. All of the consultant paediatricians and rheumatologists in the Republic of Ireland and a paediatric rheumatologist in Northern Ireland were surveyed to ascertain how many young people with JIA they were caring for in the Republic of Ireland.

4.8.1 Method for the Consultant Survey
A database of the young people in Ireland with juvenile idiopathic arthritis (JIA) did not exist, nor was there a database of the physicians caring for the group.

Informal information from clinicians indicated that only a small number of consultant physicians in Ireland had a particular interest in juvenile arthritis. The population of young people in Ireland with juvenile idiopathic arthritis was expected to be small, but was unknown. Early in the study process, the issues and challenges of researching a small vulnerable population were considered at research project meetings. Accessing the population, recruiting the group, tool development and organizational ‘gate-keeping’ practices (Silverman 2010) were all issues of concern.

The purpose of the consultant survey was to survey the total population of Consultant Paediatricians and Rheumatologists in the Republic of Ireland to ascertain (See Appendix X).
a) If they had any patients with juvenile idiopathic arthritis  
b) How many patients they had with juvenile idiopathic arthritis  
c) Within what age categories did their patients with JIA belong

The data collected would provide demographics on the number of consultants caring for young people with juvenile idiopathic arthritis and an estimate of the number of young people with JIA.

The aim was to target the total population of paediatricians and rheumatologists in the Republic of Ireland. Access to the contact details of the consultants in Ireland was gained from the National University of Ireland, Galway library copy of the current Directory of Medical Consultants in Ireland (2003) and (2004). This Directory provided details of consultants registered with the Irish Medical Association.

There were no paediatric rheumatologist consultants listed in the Republic of Ireland in the Directory. However, it was known that a paediatric rheumatologist was working in Northern Ireland. To ascertain if this consultant was caring for any young people resident in the Republic of Ireland, this consultant was surveyed also. The survey document was accompanied by an introductory letter endorsed by local consultants who were consulted on this area of the project.

The postal survey was administered at the end of August 2004 and by the end of three months the response rate was 77%. One mail shot of 146 letters was sent, this included 113 paediatricians, 32 consultant rheumatologists and one letter to the paediatric rheumatologist in Northern Ireland.
4.9 PHASE 2: Methods for Interviews with Young People with JIA to Develop Questions for a Health Services Questionnaire

**Phase 2 Introduction**

Ten young people with JIA were interviewed. The aims of the interviews were:
To provide an opportunity for young people with arthritis to discuss issues related to their healthcare services, and to form the basis of a questionnaire by generating themes and questions for a Health Care Services Questionnaire.

**4.9.1 Methods for Interviews with Young People with JIA**

The aim of this section of the project was to interview young people with juvenile idiopathic arthritis. The rationale for the interviews was to assist in generating themes and questions for the healthcare services section of the quality of life questionnaire, which was to be distributed to the targeted population in the absence of any previously designed instrument. Following ethical approval of the study, one consultant rheumatologist on the western seaboard of Ireland provided the initial database of ten young people with juvenile idiopathic arthritis to contact.

A letter from the consultant was sent to the parents of the ten young people with JIA aged 12-18. Information was provided about the project and their participation in the research was requested. A return form including appointment arrangements, contact details and a stamped addressed envelope
was provided. Appointments were made to suit the participants both in time and in place.

4.9.2 Interviews

The nature of interviews is such that a conversational attentive approach is used. Probing questions and clarification may be sought but comments and judgements are not offered (Ryan, Coughlan & Ryan 2009). The interviewer needs to provide a non-threatening environment and needs to actively listen to the interviewee using appropriate verbal and non-verbal communication. Repeating or echoing can assist with clarification of the issues and demonstrating the attentiveness of the interviewer. An advantage of interviews is that they allow for flexibility, in-depth exploration and clarification of issues.

Disadvantages of interviews include that often, samples sizes are small, time can be an issue and they are demanding of the skills of the interviewer. There is also the potential for bias (Holloway & Wheeler, 2002; Beck & Polit 2004; Tod 2006; Burns and Grove, 2011).

4.9.3 Choosing Semi-Structured Interviews

There are three principal types of interviews: structured interviews, unstructured interviews and semi-structured interviews. Structured interviews are, as the name suggests, very structured and are frequently like spoken questionnaires in order to elicit a standardised response. They provide the interviewee very little opportunity or latitude to deviate from the specified question. Unstructured interviews provide greater latitude to the interviewee to discuss the topic and the unlimited opportunity to digress from the specific topic (Parahoo, 1997; Beck & Polit 2004; Ryan, Coughlan & Cronin 2009).

Semi-structured interviews are the most commonly used interviews. The researcher normally has a set of questions or areas of interest that they wish to consider to guide them during the interview. Semi-structured interviews
provide flexibility to be responsive to the interviewee and the capacity to pursue and develop areas of interest that have been raised in the interview, while at the same time control of the interview is still in the interviewer's hands. The interviewer can also ask probe questions to pursue lines of interest and to increase the richness of the data (Burnard, 2005).

The interview guide helps to ensure that the areas of interest are covered during the interview (Bryman 2008; Silverman 2010). For these reasons a decision was made to choose semi-structured interviews for interviews with the young people, parents and clinicians in this study (please see Appendix 7).

4.9.4 Purposive Sampling
Purposive sampling was used in the study to reach a particular group of young people who had juvenile idiopathic arthritis who would discuss their health care service provision. ‘A purposive sample is one where people from a pre-specified group are purposely sought out and sampled’ (Gerrish & Lacey, 2006:182).

4.9.5 The Interview Process
Ten interviews took place. Eight interviews took place in the participants' homes. The kitchen was the usual venue for the interviews and with few exceptions parents stayed in the room. The two exceptions were interviews with young people aged 16 and 18 respectively. These two interviews were done in the living room of their home; their parents were in the house during the interviews. Two interviews were not done at home. One parent and their child came to meet me at a location that suited both parties and one interview was with a young person who was aged 18 and working, who agreed to be interviewed in the foyer of a hotel. Consent and assent were gained from both parent/guardian and the young person.

Interview preparation included preparing the questions on topics, sequencing the questions and also the prompt questions. Prompt questions are used to engage or encourage the interviewee or to assist them if they lose their train of
thought. They can also assist in gaining a balanced interview exploring issues at the appropriate depth and breadth (Legard et al., 2003). In this situation the semi-structured approach provided a framework for the interview and the prompt questions provided some leverage to broaden the discussion by asking follow-up questions.

A broad question was used at the beginning of the interview to allow the interviewee to provide an overview of a situation. This was probed further and a focused question then followed to elicit greater detail. Timing of questions is also important, as an ill-timed question can ruin an interview. The importance of the process of the interview needs to be emphasized - the introduction, the warm-up questions and the interview questions, active listening, followed by wind-down and close of the interview (Legard et al., 2003; Bryman 2008; Silverman 2010). Managing the environment and the interview are also part of the interviewer’s remit.

During the interviews, within the context of the responses the follow-up questions assisted in clarifying and deepening understanding of the issues and themes pursued. This allowed new themes to be pursued in order to gain the maximum amount of information and increase the richness of the data (Burnard, 2005). Questions such as, ‘how are your health services for you?’ led on to a discussion of GP care, hospital inpatient and outpatient care, travel time and medications and the side effects of medication and the impact of all of these on leisure time and schooling. ‘How frequently do you have to go to hospital?’ allowed for the acuity of the disease during the past year and its impact on the young person to be discussed. It also led on to a discussion on the healthcare personnel that the young people were in contact with and their competence.

4.9.6 The Interviewees

It was important to give the young people time to answer, not to anticipate their answers and to actively listen to their views (Roberts, 2000). Mindful that these young people had a chronic disease, verbal and non-verbal cues such as fatigue,
disinterest or distress were observed for (National Disability Association, 2008). Observing body language and having eye contact can assist in the interpretation of information and emotions, allowing the interviewer to appraise the situation and to react appropriately, for example recognising when it is safe to probe the participant to further explore an issue or when to wind down or terminate an interview. The young people demonstrated interest and were thoughtful in their answers. Following the interview, the interviewer was available to check if the interviewee was in a safe situation and contact details were checked before leaving collection process when particular issues relevant to those phases are discussed.
4.10 PHASE 3: Methods for Questionnaire to Young People

**Figure 4.3: Phase Three**

**Phase 3 Introduction**

The DISABKIDS Health-Related Quality of Life Questionnaire and the Health Care Services questionnaire were administered at consultant outpatient clinics on five sites throughout the Republic of Ireland to young people with arthritis. The DISABKIDS questionnaire has a generic module and a condition-specific module. For comparative purposes young people with arthritis completing the DISABKIDS questionnaire were requested to ask a peer to complete a questionnaire with generic questions only.

4.10.1 Questionnaire to Young People

Phase three of the project was the distribution of a questionnaire to young people with the agreed criteria at outpatient clinics. The instrument used was a survey questionnaire, and aimed to gather information from a population and provide data relating to specific variables. In this study the variables were to be used for descriptive purposes.

4.10.2 Principal Investigators at Research Sites

Data from the preliminary survey to the consultants indicated six consultants with a particular interest in the area of juvenile idiopathic arthritis, five of them in the Republic of Ireland. These five consultants were contacted and ethical approval sought from their institution for the research to be carried out was gained. These consultants agreed to be principal investigators for their own
institutions. Consultant public outpatient clinics were normally held on a monthly basis. This required travelling to these clinics and distributing the questionnaires. The researcher was the only person involved in the distribution of the questionnaire. This provided the work with consistency and provided me with an overview of the project and a good understanding of the nuances of each outpatient clinic.

It was important to meet with the consultant prior to each clinic commencing and check with them which patients to approach. The outpatient clinics provided situational circumstances and ‘on the spot’ decisions to be made. For example, observing patients and their families as they entered the clinic, there were instances when it was obvious that a patient I was expecting to approach to take part in the research was not feeling very well. A decision would be made not to approach the family on that day but to approach the family at another time.

The purpose and the rationale for the research were explained. An information leaflet accompanied each questionnaire (please see Appendix 8). Having given their written assent and gained their parental written consent the young people were asked to complete the questionnaire and return it. This gave the young people an ‘opt out’ situation if they did not wish to complete the questionnaire. It also meant that they were not under any duress to participate in the research. The self-assessment questionnaire provided a ‘Likert’ response scale of five categories of evaluation. For example, question one asked, ‘In general, how would you say your health is?’ The choice of response included, excellent, very good, good, fair and poor.

4.10.3 Peer Group for Comparison

The young people were asked to ask a friend of approximately the same age to complete a generic questionnaire. This was to be used for comparative purposes. The young people found this part very interesting and were full of
questions as to whom they could ask to complete the questionnaire. Parental consent and assent from this group of young people were also gained.

The European Group DISABKIDS instrument self-assessment questionnaire was the tool that was utilized. This is a health-related condition-specific questionnaire. It included health service questions generated from the interviews with young people who have arthritis, and also a generic quality of life questionnaire to a peer group without arthritis. The generic questions were embedded in the disease-specific arthritis questionnaire. The same questions were answered by their peers. The rationale for this comparison was to examine if there were significant differences between the perceived quality of life of young people with arthritis and the peer group without arthritis. This tool was chosen for a number of reasons, including, its user-friendly ease of completion. The designers of the instrument in Germany and Scotland were very accessible for discussion relating to the questionnaire and provided helpful assistance with suggestions for its distribution. The health service section of the questionnaire was particular to the health services in Ireland. These were additional questions on the questionnaire. This section was discussed with the designers of the questionnaire in Germany and this was assistive in providing external validity.

The three domains of the DISABKIDS health related quality of life questionnaire administered to the young people were mental, social and physical. Each domain had two dimensions: mental was associated with independence and emotion, the social domain had the two dimensions of inclusion and exclusion, the physical domain had the two dimensions of limitation and medication. These domains, dimensions and related subject areas served as the basis of the interviews with parents and clinicians of the young people.

4.10.4 DISABKIDS Condition Specific Module

The questionnaire probes areas relating to the impact of the disease and the understanding and appreciation that parents, friends and teachers have of the
symptoms of the disease. The diagrams below illustrate the domains and facets of the DISABKIDs health-related quality of life questionnaire that was used.
<table>
<thead>
<tr>
<th>Domain</th>
<th>Facet</th>
<th>Concept/Content</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mental</td>
<td>Independence</td>
<td>Confidence about future, living without impairments caused by condition</td>
</tr>
<tr>
<td></td>
<td>Emotion</td>
<td>Emotional worries, concerns, anger, problems because of the condition</td>
</tr>
<tr>
<td>Social</td>
<td>Social inclusion</td>
<td>Understanding of others, positive social relationships</td>
</tr>
<tr>
<td></td>
<td>Social exclusion</td>
<td>Stigma, feeling left out</td>
</tr>
<tr>
<td>Physical</td>
<td>Limitation</td>
<td>Functional limitations, perceived health status, difficulties with sleeping</td>
</tr>
<tr>
<td>HRQOL</td>
<td>Treatment</td>
<td>Perceived impact of taking medication, receiving injections, taking insulin, applying cortisone, etc.</td>
</tr>
</tbody>
</table>

**Figure 4.4: Adapted from the DISABKIDs Group (2001)**
Domains, facets and concepts/content of DISABKIDs questionnaire

4.10.5 Descriptive Statistical Tests

Descriptive statistical tests were to be carried out to explore relationships among variables. Chi-square tests were carried out however the sample numbers were too small to provide statistically significant information. The data were condensed to provide dichotomous data. This enabled the contrasting of the two groups of young people, the first group with arthritis (the cases) is contrasted against the peer group without arthritis (the control group). An odds ratio test was done. The terms odds and odds ratio are explained. Grimes and Schulz (2008:423) state that ‘odds are the probability of an event occurring divided by the probability of the event not occurring. An odds ratio is the odds of the event in one group for example those with arthritis (cases) divided by the odds in another group without arthritis (the controls) (Higgins & Green 2011). Odds ratio values range from zero to infinity (Grimes & Schulz 2008). Tabachnick and Fidell (2007:461) state that the odds ratio is ‘the change in odds of being in one of the categories of outcome when the value of a predictor increases’ by one unit. The range of values provides a 95 per cent confidence level of the true value of the odds ratio. Confidence intervals are affected by the size of the sample with small samples having wide confidence.
intervals and smaller confidence intervals with large samples. Greater confidence in the accuracy of the value is attributed to smaller confidence intervals (Pallant 2007).

4.11 **PHASE 4: Methods for Interviews with Parents of Young People with JIA**

**Phase 4 Introduction**

A sub-set of young people with ‘moderate’ arthritis was identified by a consultant rheumatologist. In-depth interviews with fourteen parents of this sub-set were conducted to ascertain the impact of the condition on the quality of life of the young people from a parental perspective.

The aim of these interviews was to learn more about the quality of life of young people with JIA from their parents’ perspectives. Interviews with parents were endeavouring to gain information relating to the impact of JIA and also living with a family member with JIA, in particular an adolescent with the disease. Areas of interest included the physical and psycho-social well-being of the young person, school, and the impact of medication and treatment that the young person was receiving. Other areas of interest included the impact of the young person’s illness on family quality of life - what was helpful to family quality of life and what was considered an adverse stressor on their family quality of life? Again, purposeful sampling was used with this group of key informants. Sharma (2004:331) describes a key informant as ‘a person who is well versed with the issue being examined and who is willing to share his or her
insight and information with the evaluator. Patton (2002:230) elaborates on the rationale for purposeful sampling. The logic and power of purposeful sampling lies in selecting information-rich cases for study in-depth. Information rich cases are those from which one can learn a great deal about issues of central importance to the purpose of the research.

4.11.1 Interview with Parents
Access to this group of parents was gained through a Consultant Rheumatologist on the western seaboard of Ireland.

A letter of invitation was sent out to this group of parents by the Consultant asking them if they would be willing to participate in the research. The letter provided the contact details of the researcher. Many of the parents contacted me directly or contacted the specialist nurses in the outpatients’ clinic. Arrangements were made to interview at a time and place that suited them. The interview schedule was structured to ensure that the dimensions domains, facets and concepts used in the questionnaire to the young people were incorporated into the interviews with parents to address quality of life issues (please see Appendix 9). Establishing rapport and trust were particularly important in these circumstances as these parents were caring for an ill child another factor was they had not been interviewed before. Fourteen parents were interviewed individually, either face to face or by phone. The length of interviews was on average 30-40 minutes.

The importance of reflection in this evolving process is integral to the study, reflecting ‘in action’ and reflecting ‘on action’. Reflecting ‘in action’ refers to the dynamics of the interview, the verbal and non-verbal cues, the need to probe or explore in greater depth and note the emergence of new themes or the repetition of emergent themes (Schon, 1983).

After each interview, review time was included as part of the process to consider the interview in a global sense, new themes, concepts, the impact of the
interview on the person being interviewed, researcher technique and affect were also considered (Dearnley, 2005).

Researcher sensitivity needs to be considered throughout the project and on multiple levels (Etherington, 2004). This is assisted by integrating an open and collaborative approach to the study and by ongoing review of methodological decisions and their consequences (Allen, 2004). In this study ongoing discussions with my supervisor provided opportunities to discuss analyze and clarify issues.

4.11.2 Telephone Interviews Versus Face-to-Face Interviews

Face-to-face interviews have many advantages over telephone interviews. Telephone interviews, however, may be cheaper, more convenient and less threatening to an interviewee. Potential participants may find it easier to refuse to participate in the research. Telephone interviews can assist in reaching participants who are geographically scattered and are possibly inadequately represented (Silverman 2010). Some writers suggest that eliminating visual contact and increasing anonymity can be very helpful in accessing participants for research project work (Cooper, Jorgensen and Merritt, 2003). It can provide ease of access and may perhaps suit the participant better. In this study interviewee preference was prioritized.

A decision was made to do as many face-to-face interviews as possible. However, interviewee preferences and pragmatism were considerations also.

The feminist literature has a strong history of discussing the importance of building rapport with interview participants (Oakley, 1981; Finch, 1984; Reinharz, 1992; Baylis et al., 1998). Non-verbal cues such as smiling and nodding as appropriate, actively listening, and managing the environment are part of the interviewer’s remit.
Patton (2002) provides information relating to the types of questions that are useful for interviewing to optimize the amount and quality of information to be gained. Different questioning perspectives can also elicit richer information. Using these different perspectives, as interpreted by the researcher, some examples are given below of questions used during the interviews with parents.

Can you tell me about your child’s arthritis? - (Knowledge question)

In the light of your experience, can you tell me more about the impact of juvenile arthritis on your child? –

(Question related to experience and behaviour).

How do you feel about that? –

(Feeling question)

What do you think about you child’s quality of life? –

(Opinion and value question).

When your child is having a ‘good day’ what do you see? -

(Sensory question).

Background and demographic questions.

4.11.3 The Interviewing Process

Without exception, all of the men who agreed to be interviewed advised me that they were not sure that they could be of assistance. However, having been reassured that the male perspective was very important to the project, and when they realized that they could set the date and time of the interview, they could not have been more helpful and indeed they did bring a different perspective to their family’s situation.

Many writers discuss various approaches to interviewing men. While it is important not to make unsubstantiated generalizations, the literature suggests...
that there are many approaches and techniques that are preferable to men rather than women. Relinquishing control of situations, for example being the interviewee instead of the interviewer, can be perceived by some men as placing oneself in a vulnerable and potentially threatening situation. Holstein and Gubrium (2003) discuss the importance of allowing symbolic expressions of control, for example allowing the interviewee the opportunity of choosing the date and time of the interview. This allows for ‘safety valves’ of being interrupted if the interviewee wants to be interrupted. All of the men in the preliminary preamble, prior to turning on the recording machinery, asked me about my interest in the research.

Probing questions and recognizing their expertise in this situation is advocated by many commentators, for example questions such as ‘Can you help me to understand?’ or ‘In the light of your experience, can you tell me more about?’ (Holstein & Gubrium 2003:60). These questions were very helpful and seemed to put the interviewees at ease; once they started talking they gained more confidence and were eager to talk about the subject. It was recognised that sensitivity was required to reduce the potential of triggering emotional trauma by discussing their child's illness and its impact on all of the family. Sensitivity and awareness to the interviewee and the topic and being able to respond appropriately were important to enable the interview to be a success and not cause ‘harm’ to the interviewee (Gorden 2003).

Interviews were tape recorded and transcribed verbatim. Data were analyzed on an ongoing basis using thematic content analysis.

Thematic analysis, also called qualitative thematic analysis was the approach used to analyze the data as it provided a useful and flexible tool for analyzing exploratory data (Schwandt, 2007). Braun & Clarke's (2006) work demonstrating a step by step six phase process of thematic analysis provided an accessible and flexible guide to analysis of the data set. (please see Appendix 10) Phase one of the process emphasizes the importance of the researcher
becoming familiar with the data through reading and re-reading the data. Phase two is the initial coding of data when many potential themes/patterns are included. Some of these themes/patterns may be refined further to become main themes as the process continues others may be excluded. Phase three is the collating of the coding into themes. During phase four the themes are reviewed and checked for compatibility with coded extracts. Phase five allows for further refining of themes. During this phase there is ongoing analysis providing more specific detail and greater clarity of the themes. The phase should culminate with the generation of names and definitions of each theme.
4.12 PHASE 5: Methods for Interviews with Clinicians of Young People with JIA

Phase 5 Introduction
In-depth interviews with Consultant Rheumatologists and Rheumatology Specialist Nurses were carried out. The aim of the interviews with clinicians was to collect data on the quality of life of the young people and address issues including the impact of the disease, its treatment and the provision of care for the young people.

4.12.1 Methods for Interviews with Clinicians
The aim of this section is to discuss the methods required to interview experts on their area of expertise. The main areas of interest to be explored during the interviews included the impact of the disease on the physical and psychological well-being of the young people with arthritis. Other areas included medication and treatment, and the social impact of the disease on the young people and their families. The provision of health care was also a major issue to be discussed.

Many of the issues relating to interviewing young people with JIA and their parents discussed in earlier sections are relevant also to this group of people, for example issues relating to semi-structured interviews, the need for preparation of the interview schedule, active listening by the interviewer and the use of probe questions. Purposeful sampling was used. The group of experts interviewed were known to be information-rich on technical and
contextual knowledge of the area being explored. Bogner and Menz (2005:46) state ‘an expert has technical knowledge, process and interpretation knowledge that is linked to his/her professional or job-related context’. The technical knowledge of the experts is very specific to their area of interest. They have process knowledge relating to routines and processes in which they are directly involved. The interpretation knowledge of the experts relates to the dissemination of specialized information, and being able to explain and interpret specialized information.

The rationale for interviewing the clinicians was to gain information from their knowledge-rich perspective. In this situation, prior to the interview the clinicians requested the interview schedule to be forwarded to them. Prior to commencing all of the interviews the aim of the research was discussed. The scope of the interview was explained, as was the processing of the information, for example that citations would be anonymised. The experts had exclusive knowledge and it was important to allow them to answer extensively. There was a need for the interviewer to be well prepared with a basic open interview guide related to topics but also with interjecting questions prepared (please see Appendix 10). The explorative expert interviews allowed discussion of motives and beliefs and enabled interaction during the interviews between the interviewee and the interviewer.

All of the interviews took place within the clinical setting in an available patient-examination room. This provided a neutral environment for all parties. The interviews were held before or after an out-patient clinic. The duration of the interviews was approximately 45 minutes. All of the clinicians were advised of the duration time prior to commencement of their interview. The length of the interviews was adhered to as agreed. The interviews were recorded and transcribed verbatim. Content analysis was used to analyze the data from all of the interviews. This type of inductive interpretative analysis allows patterns or themes from the data to be recognised and to emerge from the data (Patton 2002). Key words and themes that emerged from the data were used to
compare for congruence with data collected from the young people with arthritis and their parents.

<table>
<thead>
<tr>
<th>Phase</th>
<th>Description of the process</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Familiarizing yourself with your data:</td>
<td>Transcribing data (if necessary), reading and re-reading the data, noting down initial ideas.</td>
</tr>
<tr>
<td>2. Generating initial codes:</td>
<td>Coding interesting features of the data in a systematic fashion across the entire data set, collating data relevant to each code.</td>
</tr>
<tr>
<td>3. Searching for themes:</td>
<td>Collating codes into potential themes, gathering all data relevant to each potential theme.</td>
</tr>
<tr>
<td>4. Reviewing themes:</td>
<td>Checking if the themes work in relation to the coded extracts (Level 1) and the entire data set (Level 2), generating a thematic ‘map’ of the analysis</td>
</tr>
<tr>
<td>5. Defining and naming themes:</td>
<td>Ongoing analysis to refine the specifics of each theme, and the overall story the analysis tells, generating clear definitions and names for each theme.</td>
</tr>
<tr>
<td>6. Producing the report:</td>
<td>The final opportunity for analysis. Selection of vivid, compelling extract examples, final analysis of selected extracts, relating back of the analysis to the research question and literature, producing a report of the analysis.</td>
</tr>
</tbody>
</table>

**Table 4.1: Phases of Thematic Analysis**
(adapted from Braun & Clarke 2006)
*Qualitative Research in Psychology, 3:2, 77-101*
5. RESULTS

5.1 Results: Postal survey of Consultant Paediatricians and Consultant Rheumatologists

The aim of this survey was to gain an estimate of the number of young people with juvenile idiopathic arthritis the consultants had listed as patients. A postal survey of 145 consultant paediatricians and consultant rheumatologists, and one consultant paediatric rheumatologist resulted in a response from 112 consultants (Response rate: 77%). The only paediatric rheumatologist in Ireland was based in Belfast, Northern Ireland. This consultant was surveyed to ascertain how many patients she had with juvenile idiopathic arthritis resident in the Republic of Ireland. Letters were sent to 113 paediatricians; 82 paediatricians responded 31 did not respond. Thirty-two rheumatologists were surveyed; 29 responded, 3 did not respond. One consultant paediatric rheumatologist was surveyed and responded.

5.1.1 Numbers of Patients with Juvenile Idiopathic Arthritis?

Two paediatricians and three rheumatologists indicated that they had more than 20 patients with JIA. Two paediatricians, three rheumatologists and one paediatric rheumatologist responded that they had 11-20 patients with JIA. Twenty-four paediatricians and 13 rheumatologists indicated that they had 1-10 patients with JIA. Fifty-four paediatricians and 10 rheumatologists responded that they did not have any patients with juvenile idiopathic arthritis.

5.1.2 Ages of Patients with Juvenile Idiopathic Arthritis

Two paediatricians and one rheumatologist responded that they had more than 20 patients aged 12-18 years with JIA. One paediatrician and three rheumatologists indicated that they had 11-20 patients aged 12-18 years with JIA. Thirteen paediatricians, 12 rheumatologists and one paediatric rheumatologist responded that they had 1-10 patients aged 12-18 years with JIA. Sixty six paediatricians and 13 rheumatologists indicated that they did not have any patients aged 12-18 years with juvenile idiopathic arthritis.
Of the 112 consultants who responded, 79 (70%) had no 12-18-year-old patients with JIA: 66 (80%) paediatricians and 13 (45%) rheumatologists. Within the remaining 33 consultants, 26 (23%) had between one and ten patients: 13 (16%) paediatricians, 12 (41%) rheumatologists and one paediatric rheumatologist. Only four consultants had between 11 and 20 patients, three of whom were rheumatologists, and only three had more than 20, two of whom were paediatricians. A detailed breakdown of results can be seen in Appendix 11.

The incidence of the disease of juvenile idiopathic arthritis is approximately 1 in 10,000, prevalence 1 in 1000 (Friswell 2004; Ravelli & Martini 2007; Beresford 2011). In Ireland in 2006 there were 280,080 young people aged 12-18 years (CSO 2006). The expected number of young people with JIA aged 12-18 was 280 approximately.

Interpreting the results of the survey a projected estimate of the minimum, maximum and mean number of young people with JIA was done. Taking the minimum numbers only, twenty-six physicians had one patient only, four physicians had 11 patients, three physicians had 20 patients with JIA aged 12-18 years. The minimum estimated number of patients with JIA aged 12-18 years was 130. The maximum estimated number was 415 patients with a mean estimated number of 273 patients with JIA aged 12-18 years.
5.2 Results of Interviews with Young People

The aim of the interviews was to generate themes to form the basis of a health services questionnaire. Three key themes emerged from the interviews: communication and information, organization and quality of service, and the treatment regimen.

**Theme 1: Communication and Information Key issues**

- Friendliness of staff
- The importance of understandable explanations
- Rapid access to professionals, for example if a flare-up occurred
- Healthcare professionals to create more public awareness about juvenile arthritis

The young people were of the view that they liked the healthcare staff to be friendly and provide them with understandable explanations, for example relating to procedures and their condition. They were also aware that if they got a flare-up or if they wanted to ask a question about something that it was important to get access to the clinicians as early as possible. The young people commented on the significance of respect, the importance of a genuine sense of care from professionals and the implications of their concerns being listened to attentively. They spoke of the fact that before they got arthritis that they did not realise that young people could get the disease. They spoke too of the invisibility and the unpredictability of the disease and the difficulties of disclosing to their friends about their condition. They were of the view that life could be easier for them at school, for example, if their teachers were more aware of the disease and its implications in a school setting. The young people felt that there was a need for healthcare professionals to create more awareness amongst the general public about juvenile arthritis.
Theme 2: Organization and Quality of Service

Key Issues

- Waiting times at clinics
- Travel times to clinics
- Co-ordinated services
- Knowledge of medical history
- Time available for consultation

The young people referred to the waiting time and crowding at clinics. The fact that they were out of school was acknowledged but school work would have to be caught up with if classes were missed, and often this would occur during their own leisure time. The young people spoke of the importance of relating to people at the clinic who knew them, and knew and understood their medical history and their particular circumstances. Another related point to being known by the staff was it avoided the need to repeat their medical history. However, after waiting for a long period sometimes consultation times were perceived as very short.

The travel time to the clinic for some of the young people was more than one hour, particularly if travelling by public transport. Travelling to clinics was also associated with fatigue and other physical difficulties. One young person aged 16 who had travelled alone on public transport to attend the clinic suggested that co-ordination of appointments would be very helpful, particularly as the walk to and from the bus stop and the outpatient buildings could be problematic when coping with a flare-up. The young people were of the view that there was a need for integrated and co-ordinated services. One example given indicated that it was not unusual that one week they would see the doctor and they would have to come back the following week to attend a physiotherapy appointment. Only expert phlebotomists should take blood from them was the predominant view. Blood samples were taken from them at the hospital outpatient clinic and it would have been useful to have had it taken when they arrived at the hospital.
clinic and as they were normally waiting a long time. It would have been helpful to have received the results prior to going home so that the doctor could amend their treatment regimen prior to leaving the hospital clinic. Lack of facilities for young people at the outpatient buildings was noted by the young people.

**Theme 3: Treatment Regimen**

**Key Issues**

- Self-injecting/intravenous therapy
- Side effects of medication
- Managing the condition.

The young people spoke of their condition and their treatment regimen. Some of them were self-injecting their medications and discussed the challenges of managing their condition, for example the stiffness, gelling, pain and gastric upset due to some medications. Others were travelling to hospital outpatient clinics for intravenous medication. They indicated that they may not feel well before or after the medication being given. The specialist nurses were very helpful in providing them with techniques to ameliorate the side effects of medications and procedures. Response time and ease of access to specialist knowledge was considered extremely important to the young people.
5.3 Results of the Questionnaire to the Young People (DISABKIDS Questionnaire)

The aim of this section is to present the results of the quality of life questionnaire answered by young people with arthritis ('cases') and compare them with the responses of their peers without arthritis ('controls'). In this section only, the data have been condensed to binary data, providing one categorical response from the group with arthritis (cases) and one categorical response from the peer group without arthritis (the control group).

Population/Participants

Twenty-nine young people, male and female, aged 12-18 years with juvenile idiopathic arthritis (JIA) returned the disease specific questionnaire. Twenty-three young people without arthritis who were the peer comparator group returned the generic questionnaire.

Basic Descriptive Data are Presented

All data results are rounded to the nearest percentage. The valid percentage response for each question has been used.

5.3.1 Physical Well-being and Health

The questions relating to this dimension are centred on health and physical well-being, level of fitness and energy. The dimension explores the capacities of the respondents, the perceived extent of energy and fitness or the extent of the respondents feeling unwell.

Health Status *Cases Controls

Respondents were asked five questions relating to their health and physical well-being during the past four weeks. The majority of young people indicated that their health was excellent, very good or good (cases: n=26 (90%); controls: n=21 (95%)). Four of the respondents indicated that their health was fair or poor (cases: n=3 (10%); controls: n=1 (4%)). The Odds Ratio (OR) is explained in this situation as the probability of an event occurring in the group with
arthritis (cases) divided by the probability of the event not occurring in the group without arthritis (control group) (Grimes and Schulz 2008). A 95% Confidence Interval (CI), values ranged from 0.25 to 27.11. The Odds Ratio of 2.61 was within the range of the CI consequently the findings were not statistically significant.

**Fit and Well *Cases Controls**

Twenty-seven (93%) young people with arthritis indicated that they felt moderately/very/extremely well. There were two cases (7%) of young people indicating that they felt only slightly or not at all fit and well. Twenty-three (100%) of the peer group felt fit and well. The odds ratio was 4.61. A 95% CI, values at the lower end ranged from 0.21 to 101.03 at the upper end. The CI of the Odds Ratio (OR) spans across the null value (1.0 in this case) the respective OR is not statistically significant. The findings were not statistically significant.

**Physically Active *Cases Controls**

The majority of both cases and controls indicated that they had been ‘moderately/very/extremely’ physically active during the past month (cases: n=22, 79%; controls: n=22, 96%). One missing case for this question was noted. The OR was 0.167 using a 95% CI the values ranged from 0.019 to 1.501. The CI spans across the null value (of 1.0 in this case), the OR is not statistically significant.

**Run Well *Cases Controls**

Seven young people with arthritis (24%) indicated that they could ‘not run at all/slightly’, compared to only one person in the peer group (4%). However, the majority of both the young people with arthritis and their peer group without arthritis indicated that they could run ‘moderately/very/extremely well’. The OR was 0.143. The 95% CI ranged from 0.016, to 1.260. The findings were not statistically significant.
**Full of Energy * Cases Controls**

The majority of young people with arthritis and their comparator group indicated that they ‘very often’ felt full of energy during the past month (cases: n=24, 83%; controls: n=22, 96%) with five people with juvenile idiopathic arthritis indicating (17%) that they had ‘never’ felt full of energy during the previous four weeks. The OR was 0.218 which was within the 95% CI which ranged from 0.024, to 2.016, consequently indicating that the findings were not statistically significant.

<table>
<thead>
<tr>
<th>Question</th>
<th>Answer</th>
<th>Cases n= (%)</th>
<th>Controls n= (%)</th>
<th>Total</th>
<th>Odds Ratio</th>
<th>95%CI</th>
</tr>
</thead>
<tbody>
<tr>
<td>In general, how would you say your health is?</td>
<td>Very good</td>
<td>26 (90%)</td>
<td>21 (95%)*</td>
<td>47 (92%)</td>
<td>2.61</td>
<td>0.25, 27.11</td>
</tr>
<tr>
<td></td>
<td>Poor</td>
<td>3 (10%)</td>
<td>1 (4%)</td>
<td>4 (8%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Have you felt fit and well?</td>
<td>Very</td>
<td>27 (93%)</td>
<td>23 (100%)</td>
<td>50 (96%)</td>
<td>4.61</td>
<td>0.21, 101.03</td>
</tr>
<tr>
<td></td>
<td>Not at all</td>
<td>2 (7%)</td>
<td>0</td>
<td>2 (4%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Have you been physically active?</td>
<td>Very</td>
<td>22 (79%)*</td>
<td>22 (96%)</td>
<td>44 (86%)</td>
<td>0.167</td>
<td>0.019, 1.501</td>
</tr>
<tr>
<td></td>
<td>Not at all</td>
<td>6 (21%)</td>
<td>1 (4%)</td>
<td>7 (14%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Have you been able to run well?</td>
<td>Very</td>
<td>22 (76%)</td>
<td>22 (96%)</td>
<td>44 (85%)</td>
<td>0.143</td>
<td>0.016, 1.260</td>
</tr>
<tr>
<td></td>
<td>Not at all</td>
<td>7 (24%)</td>
<td>1 (4%)</td>
<td>8 (15%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Have you felt full of energy?</td>
<td>Very often</td>
<td>24 (83%)</td>
<td>22 (96%)</td>
<td>46 (88%)</td>
<td>0.218</td>
<td>0.024, 2.016</td>
</tr>
<tr>
<td></td>
<td>Never</td>
<td>5 (17%)</td>
<td>1 (4%)</td>
<td>6 (11%)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

* = 1 missing case
CI = confidence interval

**Table 5.1: Health and Activity**

**Psychological Well-being**

The two questions asked reflect the positive views of the respondents in relation to their lives and explores the psychological well-being of the young person.
Life Enjoyable *Cases.Controls and Fun Cases.Controls

All of the respondents in the cases group and the controls group except one person in each group responded that their life had been ‘moderately/very/extremely’ enjoyable and fun (cases n=28, 97%; controls n=22, 96%). One person from each group responded ‘not at all/slightly’ (cases: 3%; controls: 4%). The OR was 1.273. The 95% CI ranged from 0.075, to 21.513. The OR fell within the OR span accordingly the findings were statistically insignificant.

Moods and Emotions

These questions examine the moods and emotions of the young people, revealing emotions such as loneliness and sadness. The majority of young people in both groups indicated that they were ‘quite often/very often/always’ in a good mood and seldom felt sad, bad or lonely.

Good Mood * Cases.Controls

Twenty-seven young people with arthritis (93%) indicated that they were ‘always’, ‘very often’ or ‘quite often’ in a good mood compared to 23 young people without arthritis (100%). Two of the respondents indicated that they were ‘seldom/never’ in a good mood (7%) during the past four weeks. The Odds Ratio was 4.61 The 95% Confidence Interval ranged from 0.21, to 101.03. The findings were not statistically significant.

<table>
<thead>
<tr>
<th>Question</th>
<th>Answer</th>
<th>Cases n= (%)</th>
<th>Controls n= (%)</th>
<th>Total n= 50 (96%)</th>
<th>Odds Ratio</th>
<th>95% CI</th>
</tr>
</thead>
<tbody>
<tr>
<td>Has your life been enjoyable?</td>
<td>Very</td>
<td>28 (97%)</td>
<td>22 (96%)</td>
<td>50 (96%)</td>
<td>1.273</td>
<td>0.075, 21.51</td>
</tr>
<tr>
<td></td>
<td>Not all</td>
<td>1 (3%)</td>
<td>1 (4%)</td>
<td>2 (4%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Have you been in a good mood?</td>
<td>Very often</td>
<td>27 (93%)</td>
<td>23 (100%)</td>
<td>50 (96%)</td>
<td>4.61</td>
<td>0.21, 101.03</td>
</tr>
<tr>
<td></td>
<td>Never</td>
<td>2 (7%)</td>
<td>0</td>
<td>2 (4%)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

CI = confidence interval

Table 5.2: Psychological Well-being

Fun

The young people were asked if they had had fun, 28 young people with arthritis (96.6%) answered ‘quite often/very often/always’, compared to 22
(95.7%) in the control group. The OR was 1.273. The 95% CI range was 0.075, to 21.513. As the OR fell within the CI span the findings were not statistically significant.

**Sad * Cases_Controls**
Twenty-one young people with arthritis (72%) responded that they ‘never/seldom’ felt sad. The peer group without arthritis indicated that 19 (86%) never/seldom felt sad. The OR was 2.413. The 95% CI was 0.558 to 10.439. Pursuing the theme of sadness with the question ‘have you felt so bad that you didn't want to do anything?’, 24 (84%) young people with arthritis and 22 (96%) young people without arthritis answered ‘never/seldom’ to the question posed. The OR was 4.583. The 95% CI at the lower end was 0.496 to 42.353 at the upper end. consequently was not statistically significant.

**Happy the Way You Are * Cases_Controls**
Asked if they were happy the way they were, 23 young people with arthritis (82%) answered ‘quite often/very often/always’, compared to 21 (91%) in the control group. The OR was 0.438. The 95% CI range was 0.077, to 2.504. As the OR fell within the CI span the findings were not statistically significant.
<table>
<thead>
<tr>
<th>Question</th>
<th>Answer</th>
<th>Cases n= (%)</th>
<th>Controls n= (%)</th>
<th>Total</th>
<th>Odds Ratio</th>
<th>95% CI</th>
</tr>
</thead>
<tbody>
<tr>
<td>Have you had fun?</td>
<td>Very often</td>
<td>28 (97%)</td>
<td>22 (96%)</td>
<td>50 (96%)</td>
<td>1.273</td>
<td>0.075, 21.51</td>
</tr>
<tr>
<td></td>
<td>Never</td>
<td>1 (3%)</td>
<td>1 (4%)</td>
<td>2 (4%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Have you felt sad?</td>
<td>Very often</td>
<td>8 (28%)</td>
<td>3 (14%)*</td>
<td>11 (22%)</td>
<td>2.413</td>
<td>0.558, 10.43</td>
</tr>
<tr>
<td></td>
<td>Never</td>
<td>21 (72%)</td>
<td>19 (86%)</td>
<td>40 (78%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Have you felt so bad that you didn’t want to do anything?</td>
<td>Very often</td>
<td>5 (17%)</td>
<td>1 (4%)</td>
<td>6 (11%)</td>
<td>4.583</td>
<td>0.496, 42.353</td>
</tr>
<tr>
<td></td>
<td>Never</td>
<td>24 (83%)</td>
<td>22 (96%)</td>
<td>46 (88%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Have you felt lonely?</td>
<td>Very often</td>
<td>5 (17%)</td>
<td>1 (4%)</td>
<td>6 (11%)</td>
<td>4.583</td>
<td>0.496, 42.353</td>
</tr>
<tr>
<td></td>
<td>Never</td>
<td>24 (83%)</td>
<td>22 (96%)</td>
<td>46 (88%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Have you been happy with the way you are?</td>
<td>Very often</td>
<td>23 (82%)*</td>
<td>21 (91%)</td>
<td>44 (86%)</td>
<td>0.438</td>
<td>0.077, 2.504</td>
</tr>
<tr>
<td></td>
<td>Never</td>
<td>5 (18%)</td>
<td>2 (9%)</td>
<td>7 (14%)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

* = 1 missing case  
CI = confidence interval

**Table 5.3: Moods and Emotions**

**Had Enough Time for Yourself and Able to Do the Things You Want to Do in Your Free Time *Cases_Controls**

The majority of young people with arthritis (n=27, 93%) and without (n=22, 96%) indicated that they had enough time for themselves and most had been able to do the things they wanted to do in their free time 'quite often/very often/always' (cases: n=23, 82%; controls: n=20, 87%). The OR was 0.614. The Confidence Interval range was 0.052 to 7.223. The OR was within the CI range, accordingly the findings were not statistically significant.
Chapter 5: Results

<table>
<thead>
<tr>
<th>Question</th>
<th>Answer</th>
<th>Cases n= (%)</th>
<th>Controls n= (%)</th>
<th>Total</th>
<th>Odds Ratio</th>
<th>95% CI</th>
</tr>
</thead>
<tbody>
<tr>
<td>Have you had enough time for yourself?</td>
<td>Very often</td>
<td>27 (93%)</td>
<td>22 (96%)</td>
<td>49 (94%)</td>
<td>0.614</td>
<td>0.052, 7.223</td>
</tr>
<tr>
<td></td>
<td>Never</td>
<td>2 (7%)</td>
<td>1 (4%)</td>
<td>3 (6%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Have you been able to do the things you want to do in your free time?</td>
<td>Very often</td>
<td>23 (82%)*</td>
<td>20 (87%)</td>
<td>43 (84%)</td>
<td>0.690</td>
<td>0.146, 3.257</td>
</tr>
<tr>
<td></td>
<td>Never</td>
<td>5 (18%)</td>
<td>3 (13%)</td>
<td>8 (16%)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
* = 1 missing case  
CI = confidence interval

Table 5.4: Leisure Time

**Family and Home Life**

*Parents have Enough Time for You, Treated You Fairly and You Were Able to Talk to Your Parents * Cases Controls*

The majority of young people with and without arthritis considered that their parents had enough time for them, they were able to talk to their parents and they were treated fairly by their parents.

Twenty-nine young people with arthritis indicated that their parent(s) ‘quite often/very often/always’ had time for them (100%), while 22 of the control group indicated that their parent(s) ‘quite often/very often/always’ had time for them (96%). OR was 0.24 within the 95%CI range 0.01, to 6.26, thus not statistically significant. The majority of both groups (cases: n=28, 97%; controls: n=22, 96%) indicated that their parents treated them fairly ‘quite often/very often/always’. The OR was 1.273. The 95% CI interval was 0.075, to 21.513. The OR was within the CI range consequently was statistically insignificant. The young people stated that they were able to talk to their parents when they wanted to ‘quite often/very often/always’ (cases: n=27, 93%; controls: n=22, 96%). The OR was 0.614. The 95% CI range was 0.052, to 7.223. The OR was within the CI range and consequently was statistically insignificant.
### Chapter 5: Results

#### Table 5.5: Time with Parents

<table>
<thead>
<tr>
<th>Question</th>
<th>Answer</th>
<th>Cases n= (%)</th>
<th>Controls n= (%)</th>
<th>Total</th>
<th>Odds Ratio</th>
<th>95% CI</th>
</tr>
</thead>
<tbody>
<tr>
<td>Have your parents had enough time for you?</td>
<td>Very often</td>
<td>29 (100%)</td>
<td>22 (96%)</td>
<td>51 (98%)</td>
<td>0.24</td>
<td>0.01, 21.51</td>
</tr>
<tr>
<td></td>
<td>Never</td>
<td>0</td>
<td>1 (4%)</td>
<td>1 (2%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Have your parents treated you fairly?</td>
<td>Very often</td>
<td>28 (97%)</td>
<td>22 (96%)</td>
<td>50 (96%)</td>
<td>1.273</td>
<td>0.075, 21.51</td>
</tr>
<tr>
<td></td>
<td>Never</td>
<td>1 (3%)</td>
<td>1 (4%)</td>
<td>2 (4%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Have you been able to talk to your parent(s) when you wanted to?</td>
<td>Very often</td>
<td>27 (93%)</td>
<td>22 (96%)</td>
<td>49 (94%)</td>
<td>0.614</td>
<td>0.052, 7.223</td>
</tr>
<tr>
<td></td>
<td>Never</td>
<td>2 (7%)</td>
<td>1 (4%)</td>
<td>3 (6%)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

CI = confidence interval

**Money and Expenses *Cases Controls***

The majority of young people with and without arthritis indicated that they had enough money to do the same things as their friends (cases: n=28, 97%; controls: n=22, 96%). The OR was 1.273. The 95% CI range was 0.075, to 21.513. The OR value fell within the CI span and was consequently statistically insignificant. The young people indicated that they had enough money for their expenses (cases: n=25, 86%; controls: n=21, 91%). The OR value was 0.595. This value fell within the 95% CI range of 0.099 to 3.579, consequently the findings were statistically insignificant.

<table>
<thead>
<tr>
<th>Question</th>
<th>Answer</th>
<th>Cases n= (%)</th>
<th>Controls n= (%)</th>
<th>Total</th>
<th>Odds Ratio</th>
<th>95% CI</th>
</tr>
</thead>
<tbody>
<tr>
<td>Have you had enough money to do the same things as your friends?</td>
<td>Very often</td>
<td>28 (97%)</td>
<td>22 (96%)</td>
<td>50 (96%)</td>
<td>1.273</td>
<td>0.075, 21.51</td>
</tr>
<tr>
<td></td>
<td>Never</td>
<td>1 (3%)</td>
<td>1 (4%)</td>
<td>2 (4%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Have you had enough money for your expenses?</td>
<td>Very often</td>
<td>25 (86%)</td>
<td>21 (91%)</td>
<td>46 (88%)</td>
<td>0.595</td>
<td>0.099, 3.579</td>
</tr>
<tr>
<td></td>
<td>Never</td>
<td>4 (14%)</td>
<td>2 (9%)</td>
<td>6 (11%)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

CI = confidence interval

**Table 5.6: Finance**
**Friends: Spent Time with Friends, Fun with Friends * Cases Controls**

Twenty-six young people with arthritis (90%) and 22 young people without arthritis (96%) indicated that they had spent time with friends ‘quite often/very often/always’. The Odds Ratio value was 0.394. The 95% Confidence Interval range was 0.038, to 4.062. The OR value fell within the CI range consequently the findings were statistically insignificant.

Twenty-eight young people with arthritis (97%) and 22 (96%) of the control group responded that they ‘quite often/very often/always’ had fun with their friends. The OR value was 1.273. The 95% CI range was 0.075 to 21.513. The findings were statistically insignificant.

**Friends Helping Each Other and Relying on Each Other * Cases Controls**

Most respondents (cases: n=26, 93%; controls: n=23, 100%, OR was 4.80. The CI range 0.22 to 105.26. The result was statistically insignificant. The results indicated that they had helped their friends and had been helped by their friends ‘quite often/very often/always’ and that they had been able to rely on their friends ‘quite often/very often/always’ (cases: n=25, 86%; controls: n=22, 96%. The OR value was 0.284. The 95% CI range was 0.029, to 2.736. The result was statistically insignificant. Interestingly, four young people with arthritis and one person without arthritis indicated that they ‘seldom/never’ had been able to rely on their friends (cases: 14%; controls: 4%). One missing case to this question was noted.
### Table 5.7: Friends

#### School and Learning

**Happy at School, Getting on Well, Able to Pay Attention**

In general the young people had been happy at school and they perceived that they had got on well 'moderately/very/extremely' (cases: n=24, 86%; controls: n=23, 100%). The Odds Ratio was 0.531. The 95% Confidence Interval range was 0.408, to 0.690. The OR value was within the CI range and was consequently statistically insignificant. The majority of young people were able to pay attention ‘quite often/very often/always’ (cases: n=25, 86%; controls: n=23, 100%). The Odds Ratio was 0.521. The 95% Confidence Interval was 0.397-0.683. The OR was within the CI range and was consequently statistically insignificant. The young people indicated that they got along well with their teachers ‘quite often/very often/always (cases: n=26, 90%, controls: n=20, 87%). The Odds Ratio value was 1.300. The 95% Confidence Interval range was 0.237, to 7.139. The OR value was within the 95% CI range and consequently the findings were statistically insignificant.
### Chapter 5: Results

#### Table 5.8: School

<table>
<thead>
<tr>
<th>Question</th>
<th>Answer</th>
<th>Cases n= (%)</th>
<th>Controls n= (%)</th>
<th>Total</th>
<th>Odds Ratio</th>
<th>95% CI</th>
</tr>
</thead>
<tbody>
<tr>
<td>Have you been happy at school?</td>
<td>Very</td>
<td>24 (86%)*</td>
<td>23 (100%)</td>
<td>47 (92%)</td>
<td>0.32</td>
<td>0.52, 203.36</td>
</tr>
<tr>
<td></td>
<td>Not at all</td>
<td>4 (14%)</td>
<td>0</td>
<td>4 (8%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Have you got on well at school?</td>
<td>Very</td>
<td>26 (93%)*</td>
<td>23 (100%)</td>
<td>49 (96%)</td>
<td>4.80</td>
<td>0.22, 105.26</td>
</tr>
<tr>
<td></td>
<td>Not at all</td>
<td>2 (7%)</td>
<td>0</td>
<td>2 (4%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Have you been able to pay attention?</td>
<td>Very often</td>
<td>25 (86%)</td>
<td>23 (100%)</td>
<td>48 (92%)</td>
<td>9.84</td>
<td>0.50, 193.62</td>
</tr>
<tr>
<td></td>
<td>Never</td>
<td>4 (14%)</td>
<td>0</td>
<td>4 (8%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Have you got along well with your teachers?</td>
<td>Very often</td>
<td>26 (90%)</td>
<td>20 (87%)</td>
<td>46 (88%)</td>
<td>1.300</td>
<td>0.237, 7.139</td>
</tr>
<tr>
<td></td>
<td>Never</td>
<td>3 (10%)</td>
<td>3 (13%)</td>
<td>6 (11%)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

* = 1 missing case  
CI = confidence interval
## Chapter 5: Results

### Table 5.9: The Impact of Arthritis

<table>
<thead>
<tr>
<th>Question</th>
<th>Never/Seldom</th>
<th>Quite often</th>
<th>Very often/Always</th>
</tr>
</thead>
<tbody>
<tr>
<td>Are you confident about your future?</td>
<td>6 (20.7%)</td>
<td>5 (17.2%)</td>
<td>18 (62.1%)</td>
</tr>
<tr>
<td>Do you enjoy your life?</td>
<td>1 (3.4%)</td>
<td>9 (31.0%)</td>
<td>19 (65.5%)</td>
</tr>
<tr>
<td>Do you feel tired because of your arthritis?</td>
<td>14 (48.3%)</td>
<td>7 (24.1%)</td>
<td>8 (27.6%)</td>
</tr>
<tr>
<td>Is it difficult to sleep because of your arthritis?</td>
<td>28 (96.6%)</td>
<td>1 (3.4%)</td>
<td>-</td>
</tr>
<tr>
<td>Does your arthritis bother you when you play?</td>
<td>17 (58.6%)</td>
<td>7 (24.1%)</td>
<td>5 (17.2%)</td>
</tr>
<tr>
<td>Do you worry about your arthritis?</td>
<td>18 (62.1%)</td>
<td>6 (20.7%)</td>
<td>5 (17.2%)</td>
</tr>
<tr>
<td>Does your arthritis get you down?</td>
<td>19 (65.5%)</td>
<td>5 (17.2%)</td>
<td>5 (17.2%)</td>
</tr>
<tr>
<td>Do your teachers behave differently towards you than towards others?</td>
<td>21 (75.0%)*</td>
<td>4 (14.3%)</td>
<td>3 (10.7%)</td>
</tr>
<tr>
<td>Do you think you can do most things as well as other children?</td>
<td>4 (14.3%)*</td>
<td>10 (35.7%)</td>
<td>14 (50.0%)</td>
</tr>
<tr>
<td>Do your friends enjoy being with you?</td>
<td>1 (3.7%)**</td>
<td>3 (11.1%)</td>
<td>23 (38.3%)</td>
</tr>
<tr>
<td>Do you find it easy to talk about arthritis to other people?</td>
<td>9 (32.1%)</td>
<td>9 (32.1%)</td>
<td>10 (35.7%)</td>
</tr>
</tbody>
</table>

*=case missing ** = 2 cases missing

Table 5.9: The Impact of Arthritis
**Quality of Life: The Impact of Arthritis**

**Confident About the Future and Enjoying Life**

The majority of young people indicated that they were confident about the future ‘very often/always’ (n=18, 62.1%) and enjoyed life ‘very often/always’ (n=19, 65.5%).

**Physical Symptoms Related to Arthritis**

Only one young person indicated that their sleep was affected by the disease ‘quite often’ (3.4%). Fifteen young people indicated that they had fatigue ‘quite often’ or ‘very often/always’ (51.7%) compared to 48.3% (n=14) who indicated that they ‘never/seldom’ experienced tiredness due to their arthritis. Seventeen young people (58.6%) were of the view that their arthritis did not bother them when at play.

**Psychological Concerns**

Nineteen young people (65.5%) stated that their arthritis ‘never/seldom’ got them down and 18 indicated that they ‘seldom/never’ worried about their arthritis. However, 11 young people (37.9%) stated that they ‘quite often’ or ‘very often/always’ worried about their arthritis.

**Understanding by Others**

Four questions relating to the understanding of the symptoms of the disease by associated others provided interesting views. Twenty-one of twenty-eight respondents indicated that their teachers ‘never/seldom’ behaved differently towards them than others. Twenty-four young people suggested that ‘quite often’ or ‘very often/always’ they could do most things as well as other children. Nineteen young people (67.8%) indicated that they found it easy to talk about their arthritis to other people. Only one person was of the view that their friends ‘never/seldom’ enjoyed being with them.
## Quality of Life: Medical Treatment

<table>
<thead>
<tr>
<th></th>
<th>Never/Seldom n= (%)</th>
<th>Quite often n= (%)</th>
<th>Very often/Always n= (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Is it annoying for you to have to remember your medication?</td>
<td>13 (48.1%)**</td>
<td>6 (22.2%)</td>
<td>8 (29.6%)</td>
</tr>
<tr>
<td>Are you worried about your medication?</td>
<td>16 (59.3%)**</td>
<td>6 (22.2%)</td>
<td>5 (18.5%)</td>
</tr>
<tr>
<td>Does taking medication bother you?</td>
<td>14 (51.9%)**</td>
<td>6 (22.2%)</td>
<td>7 (25.9%)</td>
</tr>
<tr>
<td>Does taking medication disrupt everyday life?</td>
<td>24 (88.9%)*</td>
<td>1 (3.7%)</td>
<td>2 (7.4%)</td>
</tr>
</tbody>
</table>

* = 1 missing case ** = 2 missing cases

Table 5.10: Quality of Life: Medical Treatment
**Medication**

Fourteen young people indicated that remembering to take their medication was annoying ‘quite often’ (n=6, 22.2%) or ‘very often/always’ (n=8, 29.6%). Thirteen young people (48.1%) indicated that it was ‘never/seldom’ annoying to remember to take their medication. Anxiety relating to the medication was a significant issue. Taking the medication did not appear to bother the majority of young people (n=14, 51.9%). However, thirteen young people (48.1%) indicated that it was bothersome ‘quite often’ or ‘very often/always’. Neither did the majority of young people (n=24, 88.9%) perceive that it disrupted their everyday lives.

<table>
<thead>
<tr>
<th>In the last year . . .</th>
<th>Never n= (%)</th>
<th>A few times N= (%)</th>
<th>Every month n= (%)</th>
<th>Every week n= (%)</th>
<th>Daily n= (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>How often did you have problems with your arthritis?</td>
<td>1 (3.4%)</td>
<td>14 (48.3%)</td>
<td>2 (6.9%)</td>
<td>3 (10.3%)</td>
<td>9 (31.0%)</td>
</tr>
<tr>
<td>How severe was your arthritis during the last year?</td>
<td>4 (13.8%)</td>
<td>9 (31.0%)</td>
<td>5 (17.2%)</td>
<td>5 (17.2%)</td>
<td>6 (20.7%)</td>
</tr>
<tr>
<td>How often did you have pain in your joints or muscles?</td>
<td>1 (3.4%)</td>
<td>12 (41.4%)</td>
<td>7 (24.1%)</td>
<td>3 (10.3%)</td>
<td>6 (20.7%)</td>
</tr>
</tbody>
</table>

**Table 5.11: Questionnaire: Quality of Life (Part C) About Symptoms of the Disease**


**About Symptoms of the Disease**

Twenty eight (96.6%) out of 29 young people indicated that they had arthritis symptoms during the last year with 16 (55.1%) indicating that the severity of arthritis was ‘moderately severe/quite a bit/extremely severe’. Sixteen (55.1%) young people indicated that the frequency of pain in their joints or muscles was ‘quite often/very often/always’, with 13 answering that they ‘never/seldom’ or ‘a few times’ had pain.
### Table 5.12: Questionnaire: Part E About Your Healthcare Services

<table>
<thead>
<tr>
<th>Question</th>
<th>Excellent</th>
<th>Very good</th>
<th>Good</th>
<th>Fair</th>
<th>Poor</th>
</tr>
</thead>
<tbody>
<tr>
<td>In general, how would you say your healthcare services are?</td>
<td>7 (24.1%)</td>
<td>11 (38.0%)</td>
<td>10 (34.4%)</td>
<td>0</td>
<td>1 (3.4%)</td>
</tr>
<tr>
<td>Does your outpatient clinic doctor listen to your views?</td>
<td>0 (3.4%)</td>
<td>1 (10.3%)</td>
<td>3 (11.1%)</td>
<td>5 (17.2%)</td>
<td>20 (69.0%)</td>
</tr>
<tr>
<td>Does your GP listen to your views?</td>
<td>2 (7.4%)</td>
<td>1 (3.7%)</td>
<td>3 (11.1%)</td>
<td>3 (11.1%)</td>
<td>18 (66.6%)</td>
</tr>
<tr>
<td>Do you have to wait more than two hours at routine outpatient clinics?</td>
<td>10 (34.4%)</td>
<td>9 (31.0%)</td>
<td>5 (17.2%)</td>
<td>3 (10.3%)</td>
<td>2 (6.8%)</td>
</tr>
<tr>
<td>Would more information about your arthritis be helpful for you?</td>
<td>2 (6.8%)</td>
<td>7 (24.1%)</td>
<td>7 (24.1%)</td>
<td>8 (27.5%)</td>
<td>5 (17.2%)</td>
</tr>
<tr>
<td>Would more information for your teachers about juvenile arthritis be helpful?</td>
<td>4 (13.7%)</td>
<td>5 (17.2%)</td>
<td>4 (13.7%)</td>
<td>6 (20.6%)</td>
<td>10 (34.4%)</td>
</tr>
<tr>
<td>Would it be helpful to meet other young people with arthritis?</td>
<td>1 (3.5%)</td>
<td>7 (25.0%)</td>
<td>4 (14.2%)</td>
<td>7 (25.0%)</td>
<td>9 (32.1%)</td>
</tr>
<tr>
<td>Would more information for your friends about juvenile arthritis be helpful?</td>
<td>7 (24.1%)</td>
<td>3 (10.3%)</td>
<td>6 (20.6%)</td>
<td>9 (31.0%)</td>
<td>4 (13.7%)</td>
</tr>
<tr>
<td>Would it be helpful if your medications were free?</td>
<td>1* (3.7%)</td>
<td>1 (3.7%)</td>
<td>0</td>
<td>10 (37.0%)</td>
<td>15 (55.5%)</td>
</tr>
<tr>
<td>Would 'on-line’ prescriptions from your doctor to your pharmacist be helpful?</td>
<td>7 (26.0%)</td>
<td>3 (11.1%)</td>
<td>1 (3.7%)</td>
<td>8 (29.6%)</td>
<td>8 (29.6%)</td>
</tr>
<tr>
<td>Do you feel that you are treated with respect by healthcare professionals?</td>
<td>1* (3.5%)</td>
<td>0</td>
<td>4 (14.2%)</td>
<td>12 (42.8%)</td>
<td>11 (39.2%)</td>
</tr>
</tbody>
</table>

* = 1 missing case  ** = 2 missing cases
Healthcare Services

Twenty-eight people (96.5%) indicated that they viewed their health services to be ‘excellent’, ‘very good’ or ‘good’ with only one person indicating that they viewed their health services in general to be ‘poor’.

Listening to Views

Two questions were posed relating to listening to the views of patients/clients. The majority of patients were of the view that both their outpatient clinic doctor (n=20, 69.0%) and their GP (n=18, 66.6%) listened to their views ‘always’.

Waiting Times

Nineteen people (65.4%) indicated that they ‘never’ or ‘seldom’ had to wait more than two hours at routine outpatient clinics. However, ten people (34.3%) indicated that their waiting time at clinics was more than two hours ‘quite often’, ‘very often’ or ‘always’.

Access to Information

Four questions relating to access to information for the young people, their teachers and others were asked. Thirteen young people (44.7%) indicated that more information about their arthritis would be ‘very’ or ‘extremely’ helpful. However, nine young people (30.9%) indicated that more information would be of help only ‘slightly’ or ‘not at all’, while seven (25.0%) suggested that it would be of moderate assistance. Information for teachers was considered by 16 (55%) to be ‘very’ or ‘extremely’ helpful, with four young people (13.7%) indicating that this would be ‘moderately’ helpful.

Meeting other people with arthritis was considered positively, with twenty young people (71.3%) indicating that this would be ‘moderately’, ‘very’ or ‘extremely’ helpful. Sixteen young people (55.0%) were of the view that more information about arthritis for their friends would be helpful ‘not at all’, ‘slightly’ or ‘moderately’.
Chapter 5: Results

Free Medications
Twenty-five young people (92.5%) indicated that it would be ‘very’ or ‘extremely’ helpful if their medications were free.

On-line Prescriptions
Sixteen young people (59.2%) were of the view that on-line prescriptions from their doctor to their pharmacist would be ‘very’ or ‘extremely’ helpful, while ten respondents (37.1%) believed that on-line prescriptions would be helpful only ‘slightly’ or ‘not at all’.

Treated with Respect
Twenty-three respondents (82.0%) felt that healthcare professionals were ‘very’ or ‘extremely’ respectful of them.

5.4 Results of Interviews with Parents
Fourteen parents were interviewed individually. All of the parents were in paid employment outside the home. Interviews were set up at times to suit the parents. Participants were asked if they would prefer a telephone or face-to-face interview. Four interviews were face-to-face interviews and ten were telephone interviews. Interviews lasted 20-40 minutes approximately.

Parents were residing in four different counties on the western seaboard of Ireland. The findings of the interviews with the parents are arranged by topic. During the interviews the parents spoke about their child’s quality of life and the impact of juvenile idiopathic arthritis. Parents spoke of the direct impact of the disease on the young person’s physical well-being and psychological well-being, on their moods and emotions, their self-perception, on their social support and peers, and the school environment.

Other quality of life issues that the parents discussed were issues where the parents acted as the young person’s advocate and in their best interests, for example sourcing the most appropriate healthcare personnel to care for their
child. The impact of the disease on the quality of life of the family and their social support was also discussed. Parents made a clear demarcation between life before their child’s confirmed diagnosis of juvenile arthritis and life after diagnosis. Following diagnosis and referral to a specialist centre, the process of trying to suppress the condition commenced. Often this required a trial of medications or combination of medications before the most effective medication regimen was arrived at, and often this continued to be problematic.

Medication was a significant issue. Parents recognised that the newer biologic medications were frequently improving the quality of life of the young people, enabling them to do the things that other young people of their age were doing. However, parents were concerned about the long-term impact of these medications. Many of the interviews followed the trajectory of the disease in chronological order, as this seemed to provide parents with a natural framework in which to discuss the situation and was not imposed by the interviewer. The findings have broadly adhered to this framework. The word ‘Anon’ replaces the name of the young person when it was mentioned. Attributions to quotes are given as numbers to preserve the anonymity of participants.

5.4.1 Quality of Life: Physical Well-being and the Impact of the Disease

Parents tried to explain the impact of the disease prior to a confirmed diagnosis. The parents spoke of the pain, stiffness and fatigue that their children were coping with and the adjustments they were making to endeavour to normalize their lives.

5.4.2 Pre-diagnosis: Prior to a Confirmed Diagnosis of Juvenile Idiopathic Arthritis

*Pre-diagnosis:* ‘We knew something was wrong for a long time, and even before he went to secondary he started getting stiffness in his legs, and then some days when he couldn’t walk very well he found walking painful and we went all round the houses. We went and had him X-rayed in
[Name of Hospital] at that stage, and that would have been oh 8, 9, years ago, and they said no, no, just growing pains, he’s growing.’

(Parent 9)

Asked if he missed much school

‘So he basically would take pain killers, and he’d hobble in (to school) and by the end of the day it’d ease you know you know you’d have to perhaps drive him to the school gates or minimize the amount of walking he did on those days, but he was still determined to go in.’

(Parent 9)

Another parent spoke of the physical impact of the disease prior to diagnosis of JIA:

**Pre-diagnosis:** ‘You know serious symptoms, of some kind of problem because he was getting not just occasional stiffness, but actually more persistent stiffness, and pain. ... Coming up to that point when he was diagnosed it was quite quickly getting more serious, and I remember the image to this day - the bottom of those steps that you just walked up he couldn’t move. He couldn’t cross to come up the steps. And you know of course your heart goes out to anyone, especially young people, to be in pain, and you know he just couldn’t move.’

(Parent 10)

5.4.3 **Post-diagnosis: After a Confirmed Diagnosis of Juvenile Idiopathic Arthritis**

**Post-diagnosis:** ‘Well it does restrict them [two children], especially now in the summer time, you know, they might, you know, want to be out playing football, or doing, you know, certain sports or whatever outside, and they really can’t – you know (sigh) because of the pain and all that
sometimes you know the fatigue and all that you know that goes with it you know. It doesn’t allow them to stay out too long, you know. And it’s – it is heart breaking at times, you know. But I’m just still you know – I’m hopeful like that we will find a cure for this eventually, you know.’

(Parent 8)

**Post-diagnosis:** It was just one day he was lying on the bed and he said ‘Mam remember my brother (name of brother)’ he said ‘he had crutches where he broke his ankle’ he said ‘Mam could you get them for me?’ and I think that really took the eyes out of me. I couldn’t believe that this child here that had his whole life ahead of him was really, and when I say an invalid, we’ll say you know what I meant we’ll say. Really he was goin’ to be I thought confined...the way he was we thought he was goin’ to end up in a wheelchair.’

(Parent 13)

The disease had an insidious onset for many of the young people; however, one parent spoke of the apparent sudden onset of the arthritis.

**Post-diagnosis:** ‘I don’t think we still got the whole picture until months later really when we kept askin’ questions, you know. They may be repetitive questions ‘cause you’re in a bit of shock yourself – mm – you don’t believe it’s happenin’ you know, and like the child looks perfectly healthy. She’s not overweight you know she’s vibrant full of vitality and life and next thing bamb! Just overnight! So there’s a lot of – mm – well there was a lot of anger at the time too I tell you the truth – a lot of anger – through ignorance or just through plain anger, you know.’

(Parent 2)

**Post-diagnosis:** ‘If they could control it at all for her you know that she was not to be in pain you know if she was able to get around alright you know. You know she was always a happy girl you know, but like it’s very
hard to get up in the morning and be happy when you’re in such pain you know. I have an awful habit of asking’ her in the morning how is she, you know, and she hates it you know. I am trying to get out of that habit. It’s very hard to get up in the morning not to say anything to her you know. We’re tryin’ to cope with it all the time you know. I think in the long run that she’s goin’ to be left with the way she is. So it’s goin’ to kill her at a young age you know. The amount of medicine she has got…’

(Parent 12)

Post-diagnosis: ‘A really bad day for him is when his joints are swollen, and that’s when he couldn’t get out of bed.’

(Parent 6)

However, when the disease had been suppressed due to medication or was in remission the young person’s quality of life was remarkably different.

‘But from once he started on the Humira and the methotrexate you could see that improvement too, and you could see Anon being more outgoing. He’d play football. He’d go swimming. You know things that a normal child would do. So in that sense he was probably deprived of those in his earlier few years until he started the Humira.’

(Parent 13)

5.4.4 Quality of Life: Psychological Well-being and the Impact of the Disease

One parent spoke of their child having been seen on many occasions by physicians, but the child’s condition remained undiagnosed. The obvious distress of the child added to considerable distress for the parents. The parent spoke of their young teenager saying to his parents that they had to find out what was wrong.
Very scary, and also we were just cross because you know Anon kept on saying to us ‘you’ve got to do something, you must’ – you know he was really upset, and kept saying ‘I can’t stand it. You gotta find out what’s wrong’. It was the frustration of trying to get it sorted. That was the main frustration really, and he was very – he was always sort of raging at us saying ‘you’ve got to sort this’, you know or ‘what’s wrong I’ve got to have this sorted.’

(Parent 9)

Parents spoke of their children trying to cope with the disease, and the importance of not leaving the young person on their own and encouraging their friends to call to the house, keeping them connected with their friends. Missing school was not just about missing education but also disconnecting them from their peers.

‘And you have to keep her in a positive frame of mind as well.’

(Parent 11)

Coming to terms with the disease and telling friends about the disease could be problematic for the young people.

‘When he went on holiday with his friends and he didn’t really want to advertise to all in front of him, but in a way he had to learn to be a bit more honest about it with his friends saying “I can do so much, but we’re going to have to get a bus now lads I can’t walk” you know. He found that very hard. He didn’t want to admit to it with his friends.’

(Parent 9)

Parents were not sure if the disease shaped their children’s personalities. Several parents made reference to their children’s coping capabilities.
‘Of all my children he’s the one that could best cope with it ’cause he has the personality, and the other two, because of what they’re interested in would find it a lot harder.’

(Parent 9)

‘He was a fantastic child throughout all his illnesses. If it was the other the older lad we’d never hear the end of it. But Anon just – unless he was very, very bad with pain – Anon would say “Ah I’m ok, I’m not too bad” and you’d know – you could see it that he was suffering.’

(Parent 13)

5.4.5 Quality of Life: the Impact of the Diagnosis Process, Physically and Psychologically

During the process of diagnosis children had undergone painful procedures, attended many appointments and had been affected by the attitudes and expertise (or lack of expertise) of health professionals. Parents spoke of the difficulties of the diagnosis process on their children and their families and how stressful it had been prior to a diagnosis. Parents were looking for clarity and the experts to have the answers. Answers and clarity were not always readily available. The central focus of the parents’ attention was on their ill child.

The diagnosis process, for some, may have continued for 18 months, perhaps more, before a confirmed diagnosis was made. Parents talked of their turmoil, their helplessness, the haphazard referrals of friends and family to sources of information, the unpredictability of the disease, the uncertainty that pervaded their lives. The situation was distressing, as often the young person’s condition was deteriorating.

‘Consultants just didn’t have the experience to diagnose him. So we had to take him to where we thought had the best treatment, which was to London.’

(Parent 6)
‘I suppose like if you go back to the very start of it – ah – when indeed the problem initially developed when she was 2 odd – mm – I suppose there was a period where we were extremely concerned where the diagnosis process was ongoing, and it was quite some time before – mm – a positive diagnosis as to what exactly the problem was, was made.’

(Parent 4)

‘Thinking through what happened with Anon you know the main issue we would raise was extremely tardy diagnosis – extremely late diagnosis.’

(Parent 10)

5.4.6 Quality of Life: Parents as Advocates for their Children

Parents were acting as advocates for their children, and consequently the quality of interactions and communications with healthcare personnel was very significant for parents. Communications needed to be open and honest and the attitudes of the clinical staff were very significant. On first meetings, parents endeavoured to assess the competence of the clinicians, assess if they could trust them and if they could collaborate with them. Parents were proactive about their clinical team and if they were of the view that the clinician did not suit them or was not in their view the most appropriate person for their child’s particular needs they would make the decision to go elsewhere. Parents talked of being aware of the social cues of being listened to and being heard. They had a strong need to perceive that the clinical team was ‘with them and for them’ and that they were getting the best care for their child.

Parents and clinicians spoke of the lack of electronic record keeping, which had implications for integrated and co-ordinated care. This was especially noted prior to a diagnosis being confirmed when their child was being referred to a number of physicians for evaluation. One parent described the record keeping as ‘tawdry and at best inefficient’ (Parent 6). These less than optimal
communications between the clinical care team were not reassuring for families.

A number of parents maintained their own records. They communicated their child’s history and the most recent laboratory results using their own notes. Parents were looking for clear and concise clinical pathway information. Fathers, in particular, were looking for a piece of paper to take home - an algorithm format pathway outlined from all physicians including their first point of entry into the healthcare service system, which was usually their GP. Prior to a confirmed diagnosis parents spoke about distilling and synthesising information. Often there was a disconnection between the families and the healthcare professionals. There was a sense of parents spearheading and managing this problem-saturated situation.

‘...in fairness now her mother took on quite a lot over the years and kind of has maybe fought battles more so than I have. There was a long time there where we kind of seemed to be goin’ nowhere and eventually we came across a facility that was available in Belfast rather than here and we got her up there.’

(Parent 4)

‘And frequently the doctors were writing stuff down on more note pads, backs of envelopes, and not storing the information centrally. It’s not being shared. Personally I think it’s a total disgrace in this day and age. Because it’s the patient who suffers, and that probably applies to all – mm – areas of health service, not just in this country, but it’s just absolutely ridiculous that different categories of doctors won’t talk to each other, and that they won’t share information, which is all for the benefit of the patient again. So those two things – first of all finding the right people, and then getting them to collaborate, and share the information. It’s just so time consuming, which is very hard because when you first have a child diagnosed you’re going to be very upset about it. You’re going to have to
take on which is basically a whole project, and manage part of the health service yourself, and then fight the egos of various doctors who don’t like you taking control of your own child’s condition, ‘It’s just very tiring and very upsetting, and as I said very time consuming, and totally unnecessary, which could be fixed very easily.’

(Parent 6)

‘The biggest difficulty I think is really where you’re not in the medical profession, or you’re not – medicine isn’t your forte – mm – you can sometimes be left in a limbo – things are not properly explained or not explained in layman’s terms. I think that’s a very definite issue, but I also think – mm – you know there’s a lot more information out there now so you can kind of check things on the internet or whatever.’

(Parent 4)

‘It was a big shock for us when we were told ‘Anon’ had arthritis. But the other thing is – mm – like no one was able to tell us anything about it you know. So we just had to do our best ourselves for her you know like that.’

(Parent 12)

‘The most difficult thing, and the most time consuming thing, is the fact that we have put so much effort into building up the contacts and finding the right doctors who can help. That’s just the incredible situation that there is no one – patient type manager – who can put you in touch with the right people. We had to go to many different sources just to find out where the best place to go for treatment and therapy. Where is the right podiatrist to go and see – where is the best eye surgeon and so on. There is no single co-ordinated point, and that would be one big problem, and the second leads on from that is the fact that every time you see a new doctor there is no common sharing of the patient history or the patient notes.’

(Parent 6)
5.4.7 Quality of Life: Referral to a Regional Rheumatology Centre

Once a diagnosis was confirmed and parents were confident that their child was in the appropriate care centre and was being seen by the most competent person available, only then did parents allow the locus of control to be handed over to the physician. Many families travelled considerable distances to regional centres for their medical appointments. This was seen as a well worthwhile inconvenience as not only was the physician’s medical competence very important, parents needed to be able to trust and have continuity in their relationship with the physician. It was also seen as getting the best care for their child.

Once parents considered that they had been heard, treatment options had been discussed and a reasonable plan of action had been put in place for their child, parents acknowledged that their level of anxiety lessened, if only briefly. Having returned from the regional centres to home, parents considered flexibility and response time to phone calls and e-mails to be very important. Many were initially very surprised that they had the personal number of their consultant and found this a great comfort as well as an act of great trust. Parents recognised the need for partnership in the relationship. However, in the early days of the disease parents indicated that they were frequently overwhelmed by the whole situation and on reflection realized that it took time to build up relationships with the care team.

Having received a diagnosis and been directed to a specialist team, the abnormal started to be normalized over a period of time. All of the parents spoke positively about their rheumatology care team. The specialists, either physicians or nurses, were found to be very helpful, respectful, approachable and available to them. Understanding of the families’ perspectives was appreciated.
One parent spoke of his introduction to the specialist team:

‘She said... ‘I am the Arthritis Specialist Nurse’, and d’you know what she said – she said ‘here is my mobile phone number’. I never ever heard anyone in health ever before do something like that, and we’ve never used it, but you know the notion that her commitment to the patient meant that she could be called even you know at a weekend. That there isn’t a kind of three layer set of fences between her and you like - that’s a brilliant thing.’

(Parent 10)

One parent whose child receives specialist treatment in London said:

‘By going to a paediatric rheumatologist who specialises in this condition, and who takes part in worldwide research – someone who is very involved in the whole area – his specialist subject – we feel we’re getting the best treatment possible, and the best information possible where we have total confidence. Whereas a general paediatrician maybe very valuable in their own right, but they’re not any use to us because they don’t have the experience, and can misdiagnose very easily. So we want 100 per cent confidence, and we’ve also used the Best Doctors facility through the – mm – what was BUPA, which meant we got independent opinions from recognised experts around the world, so that we could validate what our expert in London was doing was actually the best treatment available. At that time, that was very reassuring as well.’

(Parent 6)

Parents who had travelled to Northern Ireland or Britain for assessment and treatment considered that care was different in other places. Patient expectations of care and resources were different and parents appreciated the benefit of seeing how things could be done elsewhere.
'They seemed to be far more advanced in dealing with the problem up there than we were down here. In this day and age there's no reason why the standards should be any different at any part of the island.'

*(Parent 4)*

One father indicated that he made it his priority to attend outpatient clinics with his child.

'It's very, very difficult to grab this time, but it's an absolute priority, so I've always done it, and it just seemed to me it's not you know I mean my wife and I are completely you know, shoulder to shoulder in this, but it just was for continuity better to have one person going. I've got a little file on every little thing on arthritis.'

*(Parent 10)*

Reflecting on the organization of the outpatient clinic:

'There's kind of quite a bit of a queue and occasionally a bit of chaos and you know I mean the whole thing is under resourced or badly organised or whatever. So I don't know, why is it in Sweden they don't have waiting rooms?

*(Parent 10)*

### 5.4.8 Quality of Life: the Global Well-being of the Young Person

Parents spoke of the importance of the holistic and comprehensive view of the young person. One parent, discussing the importance of sufficient time at appointments, said that the paediatric rheumatologist consultant in Britain gave them a considerable amount of time and always asked pertinent questions about the young person’s health, as well as global questions on well-being, social life and education. This consultant had known the child since he was three and was seeing the child on a regular basis; consequently knowledge of the child and the family had developed. The parents of this child found this
continuum of care very important to them, as well as rapid access to this physician by e-mail, in coping with their child’s illness.

Discussing the issue of global assessment in greater detail, one parent whose rheumatology care was in Ireland said that as well as an appointment with the rheumatologist she brought her child to a local paediatrician for an annual global assessment and consultation. The parents found this paediatric global assessment and perspective very helpful in providing a comprehensive overview of the child’s health and well-being. Issues such as nutrition, growth and other general health issues are discussed at these consultations.

‘You know there’s a lot more information out there now so you can kind of check things on the internet or whatever. But sometimes that you know you’re getting information in isolation, and that can cause concern. So one thing that we’ve always tried to do with Anon – ah – was to try and have someone who kind of looked at – at her not from the arthritis perspective, but from the Anon perspective.’

(Parent 4)

Parents acknowledged the need to see the child and not just the disease. This was encapsulated by one parent:

‘Making sure that he gets the treatment, that he gets seen by the appropriate people, and that it doesn’t impact on his social life, and his school life and his life in general. So that’s the main impact, and then you have to think about the impact on everyone else – the other children.

(Parent 6)

5.4.9 Quality of Life: Diagnosis and the Impact on the Family

On reflection parents acknowledged that the time prior to and around diagnosis was a period of acute anxiety and of crisis. They were in a high state of alertness and vigilance for and about their child. Reflecting perhaps over many years,
rarely was their anxiety for their child as acute as at the time of diagnosis. Prior to their child being diagnosed with arthritis most parents did not realise that young people could get arthritis and were shocked by the diagnosis.

Parents wished that they had got the disease and not their child. Over a period of time parents adjusted to the reality of the diagnosis that not only was their child acutely ill during a flare-up, but the illness was not going to go away - their ‘child was going to be sick for a long time’ (Parent 5). The symptoms of the disease could be suppressed but the disease could not be cured.

‘I think what frightened me was when we first heard the name I went and looked it up on Google and they always say you should never do that and I got very, very upset. You know because the long-term prognosis wasn’t good and I just heard this name and I looked it up and maybe I should have been told not to do that because that was very upsetting. And I was in tears in work you know checking through and everything...I never realised that when Anon got it first that kids under twenty anyways got such a thing, you know. So that’s a minor kind of disconcertion but I suppose also that’s actually a general public ignorance that needs to be addressed.’

(Parent 9)

‘Well I mean for somebody that’s newly diagnosed it’s a major shock and my only advice is that you have to keep goin’ with life as normal as possible.’

(Parent 11)

‘You know you’d willingly go and take it, and just give it to me, and let her off you know (sigh). The actual sickness itself it’s very hard to take really watchin’ it you know you feel so helpless, and there’s no actual cures as such you know...when you discover this first, you know, it is very traumatic on everybody involved. And not so much I suppose on the child at the time. In some cases the child might be too young to understand...but as they get
older then they’re probably saying “why me?” They may not say that to you or me, but we know what’s goin’ on in their head.’

(Parent 8)

‘I’m in my fifties you know, and you feel kind of guilty about that too, you know. ‘Cause you feel, you know, I should be gettin’ something like that instead of a young child, you know’.

(Parent 14)

Interviewing parents it was very clear that a diagnosis of JIA was a shock but for some it was also a relief. Prior to a confirmed diagnosis being made, parents had been on an ‘emotional rollercoaster’, anxious and fearful, knowing that there was something seriously wrong with their child. Many parents consoled themselves by saying ‘it could be worse’. Coming to terms with the diagnosis depended on the severity of the condition and disease activity, and life experience. Parents endeavoured to come to terms with the diagnosis and get on with life.

‘It’s shockin’ and I know there are people worse off. Kids who are terminally ill and that, and me heart goes out ‘cause I – you sit back and you look at this, and it’s the helplessness.’

(Parent 11)

One parent did have rheumatoid arthritis and talked of how important it was to be positive and make little of his own severe condition to avoid negative influences affecting his child. Parents talked of feeling guilty of perhaps passing on a faulty gene (Parent 14).

5.4.10 Quality of life: The Impact of Illness on Family Life

‘Everything has changed.’

(Parent 3)
All of the mothers interviewed were in paid employment outside the home. If their child became ill this required a juggling of family arrangements, perhaps working weekend shifts or night-duty for some mothers. Fathers working near the home often took over the caring role while mothers were at work. Parents talked of living from day to day, and making tentative arrangements only, for example family holiday plans, or a family day out, in the knowledge that these may change at the last moment due to the unpredictability of the disease.

As parents began to emerge from the shock of diagnosis and as disease activity began to subside as a result of treatment, parents began to appreciate more clearly the impact of how one member of the family being ill was affecting their other children. Feelings of guilt as a result of their lack of attention to other members of the family were acknowledged. One parent spoke about one child having completed State examinations with very little support being given from their parents because the focus of attention was on the ill child. It was not until the young person with arthritis left the family home to go to college that their other children received the attention that the parents perceived was appropriate. This was echoed by other parents.

’Sō it’s a constant worry. You never, never, never stop thinking and worrying. And then of course when we sit down we start lookin’ into the future. And I mean that really (sigh) – you know it really – we start to think like “will she be able to do this? Will she be able to do that?” And that certainly – you know – I mean it’s just something that you’re livin’ with. And then I find that maybe the other children are missin’ out because you feel that you’re just totally engrossed with her at times.’

(Parent 1)

Parents realized that siblings of the young person were anxious too.

‘They do get very worried, scared for him, and they wonder what’s happening because they don’t understand why it’s happening to him, and
Parents acknowledged that travelling to medical appointments could mean that their other children were affected by parental absences.

'I think it was 2007 he started that [medication]. Then that meant goin’ to [name of hospital] I was workin’, [name of husband] was workin’ full time, it meant getting someone here – to be here we’ll say if we weren’t back in time in the evenin’. You know I mean it did affect the family life in that way.’

(Parent 13)

Juvenile idiopathic arthritis tends to be an episodic disease with periods of crisis, then periods of non-crisis. At the time of diagnosis and subsequent flare-ups, one mother spoke of:

‘...the sadness that came over the family.’

(Parent 3)

Acute periods of the disease require rapid mobilization of resources and intense involvement of the carers. One mother had to give up paid employment for at least 18 months at the time of the initial onset of the disease when her child was three years of age. She described how her complete focus was on the ill child and how it was a full-time job managing the child’s condition -learning to give the direct care, for example injections and eye drops, co-ordinating and updating the medical team, arranging transport to medical services, and arranging assessments and applying and advocating for services on her child’s behalf. The team included the rheumatology consultant, the GP, the GP’s nurse who took the blood every 4-6 weeks, the physiotherapist, the ophthalmologist
and the optometrist. There were a number of once-only referrals to exclude or to manage related problems. Parents discussed the amount of time and energy that was spent on advocating for services for their child and the impact it all had on the quality of their family life.

During the periods of non-crisis the young people can be feeling unwell with low energy and then periods of relatively good health with better energy levels. During these periods of rehabilitation the young person may not be well enough to attend a full day at school. This may have the consequence of having extra journeys to the school or making other travel arrangements if the ill child cannot be left unattended. These types of situations can impact on the time and resources of the family and inevitably impact on the quality of family life.

Parents became attuned to the disease and its episodic nature. The fatigue associated with the chronic disease of juvenile idiopathic arthritis was strongly associated with a precursor to a flare-up, throughout the flare up and into the rehabilitative phase. Parents recognised that this very low energy was part of the disease and once the young person’s energy level was beginning to improve it was an indication that the flare-up was beginning to recede. All of the parents indicated that they maintained a high level of vigilance about their child.

‘I was constantly maybe watchin’ out for him in case he’d fall, in case he’d go out without a cap, in case he’d be getting’ a cold, had he pain, that he was eatin’ well. You know all the things a mother does we’ll say that you do, but maybe more so for him. Now we tried not to kind of make him feel as if he had an illness, but deep down I suppose some of it was rubbin’ off you know that kind of a way.’

(Parent 13)

During periods of relatively good health families were able to maintain their normal activities and enjoy a sense of relief from the crisis periods.
'A good day for me is a day that I ring Anon and Anon says – you know I’d say “how are you” and she’d say “oh great”. That’s a good day. You know but in the back of your mind you’re wondering how long is it going to last for. That’s basically, but the day that’s she’s good it’s wonderful. You know, and I mean she’s an absolutely wonderful girl.’

(Parent 1)

One parent described the impact of the weekly routine of injections to his child.

‘I mean Anon hates gettin’ needles. She’s petrified of needles, and yet she’s gettin’ them and you know when the Humara started they mix 2 bottles together, and put it into a big syringe and that used to be like – we used to – and this is something we built up for 2 days in our house the tension would build, and build with myself and my wife because we knew the injection was coming. And I’m serious about tension now.’

Then when - it used to take about 3 or 4 hours to get her ready, to get her ready to take it. Again I’m not coddin’. Then, the screams and roars when she was gettin’ it. Now I wouldn’t hold her down, [name of wife] used to do it right. But you felt exhausted just hearin’ it, and again these things have to be – these are little things that have to be looked at you know.

But there was 2 days the build up, the tension started. It would start in the morning you’d be havin’ your breakfast and [name of wife] would say ‘well Anon is due her injection on Friday’ and I’d just go ‘oh f.’

I’d start and I’d see my wife gettin’ more tired, and more edgy you know buildin’ up to this because every time it was the same. And it used to just drain her, drain me, and by the time Anon had it over and done with she’d cry for about an hour, and then she was fine. That’s awful hard you know.’

(Parent 2)
Families, however, needed to be able to adapt quickly to the particular phase of the condition as this could change rapidly. Parents and families started to develop coping skills and strategies to cope with the different phases of the condition. Initial emotional turmoil and confusion was followed by a period of struggling, often a feeling of isolation. Parents then started to become more aware of the trajectory of the illness, the early signs of a flare-up, the cycle of the flare-up or acute episode.

5.4.11 Quality of Life: Medication

It could take some time to realise that the condition could not be cured, but that newer treatment regimens could significantly improve the quality of life for the majority of patients and place them in remission. Parents talked about how the medication affected their children. Often, parents knew that it had the potential of improving the quality of life of their children. However, they all had strong concerns about the long-term impact of the medication.

‘But the concern then is, you know, what are the long-term side effects of this kind of medication? What impact it might have on him in the future. We know there isn’t an alternative at the moment, but we’re obviously worried about what might happen.’

(Parent 6)

‘The methotrexate is potent as it is, but the Humira is too, the two, and that is – that is the one thing I do hope down the line that maybe it won’t affect his liver (sigh - sounds anxious) that he might have cirrhosis of the liver or something that they might detect something like that you know. That would be the main worry I’d have with the amount of medication he’s on.’

(Parent 13)
‘The amount of medicine she has got – like her liver was givin’ a problem there now and it’s back to normal.’

(Parent 12)

As their young people grew into adults concerns relating to family planning, fertility and alcohol restrictions due to the medication were raised.

‘Like if she wants to have family – the last time she was talkin’ to doctors “you have to give us at least one year’s notice Anon”, you know. And that – mm – it brings it home to you too.’

(Parent 2)

‘I’d say he has as good a quality of life as any other teenager, and there’s nothing that we know of that he can’t do because of the arthritis. Now that may change as he gets older because you can’t drink alcohol if you’re using methotrexate, and maybe in a few years time that’s going to be a problem. Other than that he’s taking part in sports, and he’s doing very well. He’s not particularly interested in sport any more, but there’s nothing stopping him as far as I’m aware.’

(Parent 6)

Young people were endeavouring to cope with their treatment regimens, for example their medication. Some of the young people were injecting themselves and taking responsibility for storage, preparation and disposal of the needles and syringes that were used. Medication could cause them to have nausea. Living with this weekly chronic problem appeared to be a matter of fact for these young people. A number of solutions were proffered to counter this problem. However, often these did not work and the young people had to cope with the nausea on a weekly basis following the medication. Some young people were receiving their medication intravenously at outpatient clinics. This was found to be good but imposed time constraints and logistical difficulties for some when at college in a different city.
5.4.12 Quality of Life: Social Support for the Family

As parents adjusted to the situation they became more knowledgeable of service provision and supports. Supportive responses from other people could help to mitigate the impact of the situation also. The family social support network was appreciated as critical to many parents. Grandparents and aunts of the young person with JIA were seen to be the most involved with families. Grandparents were often seen as supporting the parents as well as the child. No longer in paid employment, grandparents were often able to free up parents for short periods, relieving them from a hospital room or their own home when attending hospital appointments or when other children needed to be collected from events. Often grandparents were viewed as ‘touchstones’, a quiet source of unconditional support. One mother indicated that her sisters were like ‘listening posts’; if her child was having a difficult time and she was feeling low she would contact one of her sisters and that was of enormous assistance. Knowing that she had this safety valve kept her upbeat most of the time.

Other parents without extended family living near them or whose own parents were ill or had other family difficulties described how they found themselves in an isolated and lonely situation which other people couldn’t really understand. Arthritis Ireland was singled out as providing excellent information, particularly during the past few years. Parents spoke of the importance of the teachers, particularly in primary/national school, many of whom provided extra-curricular support and were also sensitive to the needs of the young person. However, once the young person went to second-level education it was recognised that this became more problematic on a day-to-day basis due to the number of teachers involved.

5.4.13 Family Support

‘I suppose the family too were good. They were very supportive you know. When I’d be upset and worried when he really got very bad and you know you always had family and [name of husband] was there, and you always had family to say ’look it, he will be fine, he will be fine’. Now they were
only sayin’ it I suppose. They didn’t know any more than us, but I know they were supportive, and they were helpful along the way, and that’.

(Parent 13)

‘I’ve got absolutely wonderful family support. Yea, you know and if you’re not feelin’ well they understand, and they would actually listen and talk.’

(Parent 1)

Parents were of the view that meeting other parents could be helpful but may not always be helpful, particularly in the early acute stages of the disease. However, they were of the view that there was a time and place and were very willing to give of their time.

‘And as I say if there was someone out there who you know wanted to sit down and have the cup of coffee for half an hour and talk it through I’d certainly be very happy to help in that manner.’

(Parent 12)

‘A recommendation for all parents is that they join a support group. Arthritis Ireland is very good. Because if they go along to any of the meetings they’ll get the chance there to hear at the presentations - they’ll hear other parents asking the same questions that affect them. So they’ll realise that they are not in a unique situation. There’s plenty of information there, and there’s a chance to go and ask other parents who may be in a more advanced stage in dealing with their child’s problem. So they can get advice, and they can get a lot of support that way.’

(Parent 6)

5.4.14 Quality of Life: Knowledge and Information

Parents, particularly mothers, developed their expertise in recognizing the early signs and symptoms of a flare-up, ensuring that early intervention took place and trying to prevent serious ‘flare-ups’ occurring.
'Well when he was first showing any kind of symptoms there wasn’t very much information available so it’s really support groups that we’ve been able to get information. So now we have a much clearer picture of what’s involved so now we can look at things with Anon if he shows the slightest sign of trouble in his joints. Whereas we know we can get him seen very quickly. We know who to go to, and generally we’re in a better position to assess whether it’s something to do with his arthritis or if it’s something else. If the child is playing sports he can get stiff or sore, and just general activities like any other child can. So having the information, which I said came from support groups, can make it a lot easier for us to be aware of what’s likely to happen, and have less impact in the house.'

(Parent 6)

Technical advances and newer treatment regimens for people with chronic diseases continue to advance. Frequency of admission to hospital has been reduced for many patients. In the case of parents of children with newly diagnosed JIA, parents are expected very quickly to participate in and be responsible for the implementation of the treatment regimen. This expectation can provide many challenges for the family.

Learning new skills, such as helping their child with physiotherapy exercises, learning the signs of a flare-up and providing comfort measures for pain and discomfort were some of the challenges. Parental ability and competence to deliver direct care to their child was spoken about by all of the fathers.

All of the fathers provided direct support and assisted with oral medications and physiotherapy. None of the fathers gave injections: fear of being incompetent, their child was a girl and it was considered inappropriate and maternal competence were some of the reasons why they did not give injections.
5.4.15 Quality of Life: Financial Resources

Referral to regional or overseas consultants for expert opinions and management can mean a considerable time away from home. Expenses are incurred and frequently there is a loss of income. Parents tried to disassociate medical visits with negative connotations for their child and associate them with more positive connotations, if possible with the promise of a treat. This could take the form of a toy when they were younger, or an item of clothing or a special event, particularly if they were travelling from a rural area to an urban area. These too added to parents’ expenses. One parent spoke of the financial impact on the family.

‘A big financial impact, which was the cost of the travel and the cost of the treatment, and so on. But even if we hadn’t had to do that, there was obviously the extra costs just travelling about making sure he gets seen, and the fact that you are working less because you’re taking time off to make sure he gets the appropriate treatment means you earn less simply because you are self-employed. So if you don’t work there’s less money.’

(Parent 6)

Depending on disease activity, if the child was ill, public transport was rarely an option because the young person found it too difficult to walk and needed door-to-door transport. Airport and railway stations were considered to be particularly difficult due to the long walkways. However, one family whose child was being assessed in the UK and tried to integrate a short break for the family at the same time, talked about going to a Theme Park where they used a wheelchair to assist the young person getting around the park. This was viewed as a useful necessity for the day but emotionally upsetting for the parents as a possible vision of the future. The humour and the convenience of the situation was not lost on the young person with JIA, who appreciated the fact that wheelchair users did not have to queue and at the end of the day he returned the wheelchair and walked away – if very slowly.
5.4.16 Quality of Life: Social Acceptance, Normalcy and Moving On in Life

Being part of the community and endeavouring to participate in community events was viewed as getting on with life and normalizing the situation. One father spoke of his reaction to his child becoming ill and of his coming to terms with his child's disease:

‘Well I think initially like you were there, and the fear was like what is this? Is she going to be dead in 6 months? Is she going to be dead in a year? And then will she be able to walk? Will she have a normal life? What will it be now – fine, we know she’s never going to run the Olympic marathon, nor the 100 metres sprint. But those are fine like you know life goes on without those. Like, there’s a very small number of people who’ll ever do those things.’

(Parent 4)

‘But you know anything short of death is still life, so we’re getting better already you know. And actually Anon’s version, I mean I don’t know but I’d have a terrible feeling that you know arthritis can be much more savage.’

(Parent 10)

‘I was an only child, but my father was actually wheelchair bound for most of my life...it could have been arthritis-related I don’t really know – mm – but, so I kind of – there’s always been a problem in the background so I suppose maybe my acceptance is a little bit easier because of that.’

(Parent 14)

Adapting to the situation one parent spoke of how their child made light of his illness with his peers.

‘I remember he – he did it very very well – he said to me, his friends, mm – “oh I can’t go away next weekend we’ve got to get you know the – mm – sorted, it seems I’ve got arthritis”, and of course their reaction to it is a bit
like the “C” word – a highly dangerous medical term – and you know he dealt with it very well by saying something “Oh I’ll probably only have one leg next time I see you, but I wouldn’t worry about that you can push the wheelchair” – this kind of stuff. He made light of it. And so the kind of potential social impact of someone in the peer group suddenly being ill, or having something different you know seemed to have not really occurred.’

(Parent 10)

Fathers particularly spoke of the need for hope in the future, the potential of new advances in treatment, of having a philosophical stance of optimism and hope, and just getting on with life and coping with the disease. Young people told their parents that they wanted to be the same as their peers and not treated differently. However, parents realized that depending on disease activity, this was clearly not always possible. Parents spoke of turning a blind eye and giving concessions because their child was having a flare-up. Adhering to treatment regimens, including exercise programmes, could be difficult.

Mothers of older teenagers at college said that they used text messages to remind their children to take medications, to get their medication from the pharmacy, and to go to appointments. The mothers said that they should not be doing this but they knew their children would forget. They recognised that this was a period of transition and this was a form of support which would gradually be reduced as the young person took over responsibility for their own care.

Parents spoke of the young person with JIA with great pride, that during adverse circumstances the young person demonstrated strong positive traits of character and was perceived as a ‘fighter’. They spoke of their strong coping abilities and how important it was to strengthen these resources.
‘It’s a serious problem you know but he managed to cope with it. We don’t have any problem coping with it. If he can cope with it we can cope with it. What are we meant to do? It’s his body.’

(Parent 10)

As the young person became an older teenager, parents or another adult still attended clinical appointments with them. Parents were very aware that transition was a process and considered they needed to support the young person at outpatient clinics. They also wanted to hear what the physician was saying.

‘And like, you know, and the thing about it she’s a fighter, which is wonderful, and I take pride in the fact that she is a fighter – like she’s in college, she’s getting first class honours, but yet – doin’ her exams, the stress comes on, the flare-ups come, and there’s one part of you sayin’ just pull her out, you know. But the other part is sayin’ – you’re watchin’ her and she wants to do it.’

(Parent 2)

‘To have coped as well that’s why I said he’s just fantastic, and anybody that ever had the – we’ll say has met him or if they know any little bit about illnesses or that, think he is just brilliant the way he has coped with all he has got at this stage in his life.’

(Parent 13)

5.4.17 Opportunity to Talk with a Researcher

Parents considered their time important. Many of them were self-employed and time was an important and valuable commodity. Speaking to a researcher was important because it could help others by creating awareness of the condition. None of the parents had ever had an opportunity to talk to a researcher about their child’s condition and its impact and welcomed the opportunity.
5.4.18 Summary of Results of Interviews with Parents

In-depth interviews with parents of young people with JIA were undertaken. Parents spoke of their shock, disbelief, fear and anxiety at the time of diagnosis but relief that the disease could be treated. Endeavouring to reduce the impact of a flare-up, and vigilance for signs of symptoms of the disease, became normal part of their daily lives. Developing contingency plans for changing contexts and circumstances to cope with this unpredictable disease also became normal for parents. Endeavouring to access resources and services and the need for coordinated care resonated with all of the parents. Recognition of the practical difficulties that children were encountering and pride in their efforts to adapt to particular situations was a major theme. Education, school services and future prospects were frequent issues.

Social support from family, friends and others was influential in assisting parents cope and adapt to the situation. Socialization as an issue was considered very important. Parents spoke of their children disguising their symptoms or ‘passing them off’, such as ‘a bit of bother with my knees’. Parents acknowledged that the newer biologic medications had improved the quality of life of their children. However, parents had strong safety concerns relating to the long-term adverse effects of these medications that their children were taking.

5.5 Results of Interviews with Consultant Rheumatologists and Rheumatology Specialist Nurses

Two rheumatologists and two specialist nurses working in the Atlantic corridor of Ireland were interviewed. All of the interviews took place in their workplace. The interviews lasted on average 40 minutes. Prior to the interviews taking place the topics scheduled for discussion were either discussed with the four clinicians or sent to them for consideration.
5.5.1 Diagnosis

At the early stages of the interviews the clinicians spoke about the disease of juvenile arthritis, how it presents and its impact on the body. Physicians spoke of how difficult the condition of juvenile idiopathic arthritis is to diagnose as it mimics other diseases. Even in the best international centres with the best facilities it may still be difficult to diagnose. Clinically, it was recognized that there was a need for a strong paediatric input to the management of the disease in children.

The clinicians also had extensive experience of caring for adults with arthritis. They indicated that the children were not ‘mini adults’ and the symptoms of the disease were different in children than in adults. So also was the treatment. Referral delays to the rheumatology service could be due to the lack of visible signs and symptoms and the lack of complaining by young children. One rheumatologist suggested that first-line physicians who were non-specialists in the area of rheumatology may not always have the proficiency to do musculoskeletal assessments in children. This delay in diagnosis could also cause a delay in referral to a specialist physician.

‘If the child isn’t limping they’re deemed to be OK. But if the child has a limp it seems to trigger off a referral, but a lot of kids don’t limp until there’s a lot of chronic change. They may have arthritis elsewhere that doesn’t get picked up.’

(Consultant)

Delay in referral can also result in potentially avoidable irreversible changes or damage to the joints occurring in the intervening period prior to being treated by a specialist. Early diagnosis can prevent the occurrence of this vista.

‘Today the way we’re able to manage diseases like arthritis the earlier the diagnosis the better for the patient, and the key is we can prevent damage, but we can’t fix it when it happens. And it’s very well documented today
particularly in adults that if we diagnose people early and we get them the right treatment early they go into remission, and they don’t have long-term disability. And that’s going to be a great gift to them for the rest of their lives. They’re going to be able to work, do whatever they want to do, and they won’t have pain and suffering.’

(Consultant)

Reactions to the diagnosis were manifold. However, shock and denial were frequently manifested. The team endeavoured to assist families to understand and cope with the situation.

‘Denial is a big factor in the early days. I find the best route helping them [parents] cope is helping them to understand the disease, to understand the disease process.’

(RSN)

‘My experience here is parents just don’t want their kid to have arthritis, and they won’t even agree that their child has arthritis sometimes, and that’s a real barrier. Because once they won’t accept the diagnosis they’re certainly not going to accept the treatment.’

(Consultant)

Clinicians acknowledged parents needed time to take the diagnosis in, have it explained and have it confirmed before they could ask questions. Avoiding overload of information at the early stages was acknowledged as very important as families had difficulties processing it. It may take a few visits to the clinic before they were able to ask probing questions.

‘They’re in shock, and it’s only when they come back the second or third time that they have the questions for you. Because the first time really is, “are you sure what they have, and but isn’t that for an old person?” And then they go away, and they ask other people and they tell them “Oh yes, I
know somebody who has it”, and they put them in contact with somebody else who has a young kid, and it’s “oh yea now I realise’."

(RSN)

Once the parents started to process the information and started to accept the situation it seemed that there was a real and searching need for information. The short-term and long-term impact of the disease, the impact of all medication and treatment modalities were scrutinized. Concerns relating to the minutiae of current daily life, to career, work and family issues in the future were raised as parents endeavoured to understand the ramifications of this disease for the young person and the extended family social network.

‘Medications, going to school, will it affect their schooling, affect them playing, doing sports, interacting with – having kids themselves? I mean that’s how far ahead they think. At the very beginning they’re thinking of their fertility and then how will they tell other kids, what should they tell them that they have arthritis they shouldn’t be playing or should they say don’t tell them that they have it, or should they tell teachers, and you know that’s mostly what they’re worried about initially.

(RSN)

The specialist nurses, recognising the difficult path that the parents and their children were negotiating, spoke of the expertise of the specialist physicians in assisting families come to terms with the situation.

‘Absolutely superb with the children, and with the parents, they ease them into it very well.’

(RSN)

The consultants considered juvenile arthritis to be potentially very serious but if managed well it was not the worst condition to have.
‘Ok, it's a painful condition in some people at some stages, but most of the time pain isn't a huge issue, and most of the time my focus would be on the future. It would be on present function that the patient can go to school, be part of the family, play their sports in order that they have the optimum education so that in adulthood you know they're starting off equal to their peers, and not as a disabled person behind their peers and needed to be specially minded.’

(Consultant)

The clinicians considered that being proactive and complying with the treatment plan would allow the young people to feel better and would reduce the impact of the disease on other aspects of the young person's life.

‘You know if Johnny has arthritis, well he does have arthritis, that's OK. The important thing is not to ignore it but to manage it. And to take a proactive step rather than running around flailing in the wind hoping it goes away on its own.’

(Consultant)

You're facilitating the child to be normal at home and at school. You are not facilitating the child by giving them notes to come off PE and notes to be off this, and can’t do that. It’s a whole pile of you know notes we don’t write. We don’t make the children disabled. We don’t give them excuses to not be playing football. If they don’t want to play football they don’t want to play football, but it’s not because of us.’

(Consultant)

Specialist physicians, acknowledging the impact of juvenile idiopathic disease in the past, were very optimistic about the future. Not only were the medications improving quality of life, there was optimism about the potential of gene therapy in the future. However, the team was concerned with adherence to medical and physiotherapy regimens in order to avoid residual problems of the disease.
'We know the down side of not getting the medicines is that the arthritis is active and you get permanent damage. We know the upside of taking the medicines you don’t have that. And as far as we know there is no long term consequence to most of these medicines and you just have to explain that. You know you can’t be absolutely certain. But there are certainties. There are certainties about not taking the medicine, that you do have long term impact...And a lot of them will get medicines that will keep them well until such time as there is a better medicine.’

(Consultant)

5.5.2 Young People
Getting to know the young people, building a rapport with them and their families and explaining to them the disease and its management and the individual plan of action for each patient were recognised as being very important by the whole team. This needed to commence from their first meeting.

‘The education starts really with the diagnosis, and meeting the patient, and you get to know your patient through doing education with them. You get to know the patient, and what they’re – what they’re capable of absorbing, and what understanding they have of the disease, and how you think they’re going to be compliant with medication.’

(RSN)

The young people did not always complain about pain or discomfort. They needed to be asked very precise questions to elicit information from them. Following an assessment for pain, an appropriate course of treatment would be recommended for the individual young person.

‘I think the thing is to assess exactly where the pain is, and what type of pain it is. And once you get a handle then of where it is and ask the questions: is it day, morning, noon is it after sport, before sport, worse on rest, worse on activity, and find out what it is, and then if you have a good
knowledge of what drugs you have available, their side effects, and then decide, well, will they be reliable taking them? Are you better off to give them one tablet to take in the morning, or is it one tablet three times a day or will they be reliable enough to take them or will it affect their schooling. Will you have to be sending tablets into school?’

(RSN)

Uveitis, an inflammation of the uvea of the eye, is associated with juvenile idiopathic arthritis. An adult with this condition would most likely be complaining of strong pain. Children may be aware of some visual disturbances but rarely do they complain of pain. Eye examinations need to be done regularly in young people with JIA, particularly certain categories of JIA, to preclude or monitor this condition to avoid serious eyesight deterioration.

‘Kids don’t complain so the commonest symptom of uveitis in kids is blindness. So that’s a problem, so that’s why you have to screen. We don’t see too much of that. It’s just picking it up early and being aware of it, and getting them on disease modifying drugs early is a kind of a key there. And thankfully we have very good ophthalmologists here and fairly quick access for the kids. So that’s a huge plus.

(Consultant)

The rheumatologists spoke warmly of their patients and acknowledged that everybody was on a ‘learning curve’ – the care team, the patients the families. Over time the young people tended to be very knowledgeable about their disease and sometimes had new ideas and suggestions about improving the care for themselves and others. They indicated also that the young people would endeavour to calm their parents and allay parental anxieties, demonstrating greater psychological maturity than their years would suggest.

‘By and large I think when kids are very proactive about their disease they do great, and like I say they tend to know more than their parents. They
end up calming their parents down, and doing very well. So it is with diabetes where the kids actually know much more than their parents know, and sometimes more than the doctors and nurses know. And that’s a good thing. I don’t think it’s a bad thing. Well I’m not challenged with people who learn more than me. But they adapt their life to it, and they figure out ways and means of doing it. Sometimes they come up with bright ideas that we can give to other kids. “You know this is actually something I did and might be helpful for other kids”, and actually these can be breakthroughs in how we manage these diseases so it’s a good thing to do.’

(Consultant)

5.5.3 Medication/Treatment
Learning about the medications, the dosages, the times to be given, the route to be given and understanding how, why and when the prescribed medication works takes time. During a flare-up, some young people may be on more than six daily oral medications and perhaps eye drops also. Being able to see immediate improvements can assist with the compliance to regimens. Some medications take longer to act as they need time to build up in the system, and consequently patients may not perceive positive results for a few months. The young people and their families needed to understand the action and interactions of the medications. Often parents were fearful about some of the medications. They had consented to the medication but reading the literature accompanying it was not always reassuring. The physicians were aware of these fears.

‘And there’s a big fear factor around drugs like methotrexate here still. It’s a lack of knowledge, and people just hear the word chemotherapy, which sets off all the alarm bells, but they don’t understand that this is a really, really good drug for kids with arthritis, and it works really, really well, and it’s much safer than many of the other medicines that are out there.’

(Consultant)
Methotrexate is a medication that is commonly used in the treatment of young people with juvenile idiopathic arthritis. It is administered frequently as a tablet, which is yellow in colour, or by a subcutaneous injection. The specialists were all very aware not only of the physical side effects of the medications but also of the psychological side effects of them.

‘They have psychologists in the States who do colour yellow desensitisation programmes because kids have phobias just with the colour yellow because of the methotrexate. So you know these are real issues that we shouldn’t ignore, and we need those services. We probably could do with some counselling services, and not just for kids but from what I see for parents.’

(Consultant)

The need to avoid or reduce the physical and psychological trauma was very important. The specialists recognised the likelihood that the young people would be on some of these medications for protracted periods. Monitoring of their blood on a very regular basis is part of the treatment. The procedure requires the use of needles to take a sample, which children can find traumatic. Young children were particularly vulnerable to being traumatized.

‘And the younger the child I think the more needles we can avoid the better they are, that we don’t traumatise them for life with needles, and shots and IVs and whatever else we do.’

(Consultant)

5.5.4 Communication

The specialist team is a small one, and consequently they get to know the young people and their families well. The specialist nurses took a very particular interest in their patients and families and were aware of important family occasions such as communions and confirmations occurring.
'The staff in Outpatients basically stay the same. So you get to know them [patients]. You get to know their families. You know when there’s a Holy Communion coming up or Confirmation.

(RSN)

The physicians acknowledged that the specialist nurses spent more time with the patients and their families and had greater potential to build up stronger personal relationships with them. Development of an understanding and rapport with families had the benefit of assisting the families to be more comfortable in asking questions. The physicians acknowledged that depending on the circumstances sometimes it was easier for the families to talk to the specialist nurses than themselves.

'Well the nurses do a lot of counselling I suppose for the adults of paediatric patients. They probably get on better with some of the families than the doctors do I would say.’ And maybe the nurses may be able to get more useful information than we’ll get.

(Consultant)

The specialist nurses noted that the parents may have accepted the diagnosis but they were always on the alert for new information about juvenile arthritis. This could be on the Internet or from friends in other parts of the world. Parents very often would contact the specialist nurses by text, to check out or verify something that they had heard or read.

'It's just amazing what they Google and everything, and looking for or talking, or they might have been in the States visiting somebody else and they’d be saying we have a new drug how would I get it – they think that Ireland is really far behind you know. “Oh no” we said, “we’re giving those but we said we’ll try these first” and then they are amazed that actually the drugs are available here, and they can be more reassured that if what
they’re on doesn’t work that they will have a choice. But reassurance is the main thing.’

(RSN)

The team agreed that the physicians tended to do more of the decision making relating to medical treatment, while the nurses spent more time with the families and had a strong interactive role with families.

‘I think we tend to do more of the decision making maybe about well this is the diagnosis, this is the medicine but there is probably more holistic interaction between parents and nurses, when you’ve a nurse specialist who can spend another 15 minutes with them where we might have spent 5 minutes. I think that’s very important, and they have a day ward here and phone service which you know brings a bit more interaction and there’s certainly a feeling for parents, “great now we can contact them if there is a problem”.’

(Consultant)

The quality of the interactions between patients, families and clinical staff is critical in the provision of appropriate care. The team were aware that at times the young people found it difficult to discuss their situation and acknowledged this could be problematic.

‘If kids don’t tell us what their problems are it’s very hard to address them, whether it’s a fear of the medicine, side effects of the medicine, worry about how their arthritis is going to do over time, fear about exams, concerns about being able to play with their friends. You know these are real concerns for kids. But sometimes the kids or their parents don’t voice these concerns, and it’s hard for us to appreciate just how much impact these things are having in their lives until they tell us that.’

(Consultant)
5.5.5 Culture

One clinician, having gained experience working in North America, spoke of some cultural differences they had noted. Parents in Ireland appeared to be intimidated by consultant physicians, while this was not their experience in North America. Also, parents in North America appeared to be more accepting of the disease and of the treatment protocols. In Ireland parents indicated that they did not want their child to have this condition and they had significant fears about the medications. This was perceived as a barrier to the child’s care.

5.5.6 Young Adults and Transition from Juvenile Services to Adult Services

All of the specialists agreed that moving from juvenile to adult healthcare services was considered a process that required sensitive management. Parents needed reassurance that the young adult had the capacity to take control of the situation and be independent. The specialists were aware that taking in information at clinics could be problematic and from a helpful resource point of view it could be useful to have another person with the young person for company and to verify information.

The team was aware that parental concerns were related to the young person lapsing into a flare-up without the usual trigger reminders regarding their medication and possible damage to joints as a long-term consequence of the flare-up. The parents needed reassurance that the young people could ‘go it alone’ successfully before they would gradually withdraw.

The specialists spoke about the very involved care of the parents over a long period and their possible sense of isolation and exclusion as a result of them not being able to provide reminder triggers for the young people as the management of the disease may have changed. This role change could be difficult for the parents to come to terms with. The young person, however, was the patient and the long-term relationship was going to be with the patient. The
overarching point made by the team was that transition to adult services was a process and should not be an abrupt departure from one service into another.

‘I think as long as they’re legally dependent on their parents I suppose the parents need to be involved. I think two heads are better than one. But yes when they become independent you have to give them the option of “do you want your Mum and Dad in? Do you want your Mum and Dad to know?” And then I suppose it’s not even medico-legally but just you know for the sake of the patient, and getting as much information from them that we can help them with. I suppose it’s important for them to have some independence, and some confidentiality too.

(Consultant)

The need for flexibility, being able to adapt and adjust to particular circumstances was considered to be part of the ‘bread and butter’ of the job of the specialist team.

‘So there are issues, and we have to be adaptable and we have to adjust the plan and the approach depending on who is in the room, and we do.’

(Consultant)

As the young people got older, new issues emerged. The team endeavoured to manage their care to allow them as normal a young adulthood as possible.

‘The transition is I suppose more difficult, not so much in their teenage years as to when they get to 19, 20, 21, and they want to go to Australia for a year, or they want to go to America for 3 months, and they have medications that they need to bring with them, or things like drugs or alcohol or having babies and these kinds of things come on board. That’s where we really start to run into a more of a – I suppose a socio-medical issue as much as anything else. Because the issues change, and the needs change, and patients become more independent, and don’t really want
their parents involved in some of these decisions as they get to young adulthood. And that can be difficult. Maybe it’s less of an issue here than in the States, but young pregnancies in High School and things are a big issue for juvenile arthritis in the States, a much bigger issue than here.

(Consultant)

Another Consultant described his experiences of the process of transition for the parents. Referring to the parents:

‘They disappear. They disappear, and they usually disappear appropriately. I like to think it’s because they have a trust that what we are doing with the child is appropriate and the child is now reaching independence…’

(Consultant)

5.5.7 The Healthcare Service for Young People with Juvenile Idiopathic Arthritis

The rheumatologists were very clear that the medical treatment of JIA had improved significantly over the years. Newer medications had reduced joint damage and disability. Some parts of the service were very good but other parts were less than optimal. Rapid access to other consultant services, for example orthopaedics, ophthalmology and laboratory services, were very good. The In-patient rheumatology service had decreased and the outpatient service, including an intravenous therapy day ward, had increased. However, there was a deficit of paediatric physiotherapy, and occupational therapy and psychology services were under-resourced. There was a great need for more education and different approaches such as group clinics for the young people.

‘As rheumatologists we know what to do. We have a stepped approach. We’re comfortable making that step up, but if we had the other resources it would make things a lot better for sure.’
‘We have a big deficit of allied health professionals here, and we have a big knowledge deficit when it comes to juvenile arthritis. I think that they’re our biggest barriers to care.’

(Consultant)

The clinicians acknowledged that the young people were not assessed for quality of life issues. Education and training in this area would be helpful. Quality of life assessments had been introduced in adult services and had proved significant.

‘Probably the one thing we fall down on with kids is we probably need to assess them a little better in terms of childhood assessments for quality of life, and how they’re managing, and how they’re coping in Clinic. We’re doing that for adults. I think it’s easier for adults because we’re trained more in adult rheumatology. But we probably could do with a bit more help, and training for that, or for similar measures for kids.’

(Consultant)

The rheumatologists spoke strongly about communications between Primary Care givers and others. Delays in communications not only between external services but also internal services were not only frustrating but they could impact on patient care. Communication services needed to be brought into the 21st century with the use of real time data and other electronic medical records communications systems. The current services could be frustrating.

‘So you don’t have to wait 6 months for your dictated letter to come out, and all these kinds of things that are 40 years out of date.’

(Consultant)

Financial control relating to the provision of services was not in the control of the clinicians. The rheumatology service could be improved according to one physician if there was more collaboration between the financial control decision
makers and the front-line decision makers, as this could result in more effective use of resources.

‘The people who actually know how to manage these diseases don’t get the credit they should do from the people who run the system. So as a result we have people who don’t know how to run an economic system making decisions about how everybody else should do their work rather than the people who know how to make it work.’

(Consultant)

5.5.8 The Role of the Team

The team were of the view that the service that was being provided was not perfect but it had a lot of positives. They acknowledged that they were a small cohesive team and they enjoyed working together. Specialist physicians were very optimistic about future treatment modalities.

Often families would know their physician or specialist nurse for many years; often they would have ‘travelled the difficult road’ with them during flare-ups of the disease or other crises related to the disease. These connections were strong.

The role of the nurse revolved around coping primarily with the patient and with the family, and linking with the consultants and other members of the allied health care team such as the physiotherapist, the Occupational Therapist and others in the multi-disciplinary group.
5.5.9 Summary

The main issues to emerge from the interviews with the specialist physicians and specialist nurses, when asked about the quality of life of young people with JIA, related to the care and management of the young people and their families. The treatment issues were often complex. Navigating the young people and their families through the disease trajectory, which was not always linear, the team encouraged the young people and their families to lead as normal a life as possible. The team endeavoured to instil an approach of acknowledgment and acceptance of the JIA, to be proactive in its management, and to enjoy life. Specialist paediatric allied health services needed greater resources. Updating communication services of medical data needed to be prioritized urgently by economic decision makers. As the young people grew into adulthood the issues changed and the problems and challenges were different.

The problems associated with JIA were mainly manageable, although some were very complex. If one medication did not work others would be tried. Clinicians associated medication efficacy with improved health status. Improved health status was aligned closely with improved functional status with a resultant improvement in quality of life. An unfortunate reality was the side-effects of medications. Minimizing trauma to young people due to invasive procedures and the side-effects of medications was given a high priority by all of the clinicians. Good communications with the team were necessary so that the management regimen could be adapted to the young person’s particular needs, and flare-ups and damage to joints and eyes could be reduced. The young people themselves often had ideas and suggestions on how to improve care for themselves and others. Learning about the disease and its management were ongoing and required continuous updating, not only by the young people and their families but also by healthcare providers including the specialist team.
Figure 5.1: Key Issues to Emerge from the Study
Findings:
Issues that emerged from exploration of the QOL of young people with arthritis study. The purpose of the diagram is to indicate the key issues of the 3 groups, Young people = , Parents = , and Clinicians = 

Figure 5.2: Demonstrating Convergence and Non-Convergence of Findings
6 DISCUSSION

6.1 Introduction

The aim of the study was to explore the health related quality of life issues of young people in Ireland with the auto-immune disease of juvenile idiopathic arthritis using a predominantly qualitative naturalistic approach. The study documented the views of young people with JIA at five locations and the views of parents and clinicians in one geographical area in Ireland. Survey results and interviews with parents, clinicians and young people provided deeper richer understanding of the context, quality and reality of the lives of young people with JIA from the perspectives of these groups (Green et al 2001; Feilzer 2010).

The incidence of the disease is approximately 1 in 10,000, and the prevalence is 1 in 1000 (Friswell 2004; Ravelli & Martini 2007; Beresford 2011). In Ireland in 2006 there were 280,080 young people aged 12-18 years (CSO 2006). Using the Central statistics office figures the estimated number of young people with JIA was 280 approximately. Interpreting the physician survey the minimum estimated number of patients with JIA aged 12-18 years was 130, the maximum estimated number was 415 patients with a mean number of 273 patients with JIA aged 12-18 years. The total number of young people to be included in the study meeting the eligibility criteria of age and a diagnosis of JIA for 2 years or more was forty-eight. This number was significantly lower than that expected. The total number of questionnaire respondents who had JIA was twenty-nine (response rate 60%). This small sample of questionnaire respondents had implications for reliability and generalizability of findings.

In the preliminary stages of the study the conceptual and philosophical frameworks underpinning, structuring and supporting the study were considered. A pragmatic approach was chosen as the method to answer most effectively the research question (Tashakkori and Teddie 2003; O’Cathain 2009). Using triangulation allowed for integration and interpretation of these findings (Jick 1979).
Many of the underpinning health promoting structures were steadfast throughout the study, for example the Ottawa Charter (1986), but the significance of other supportive concepts evolved and developed in depth and breadth during the course of the study. An example of this was the concept of health protection, with social support and resilience evolving in the study from this concept. Adaptation emerged as the central unifying dimension across the study. This included adaptation to the disease, the treatment regimen and to life with juvenile idiopathic arthritis.

Chapter six is divided into three sections.

Section one discusses findings of the study:
The impact of JIA on the bio-psychosocial well-being and quality of life on a young person with juvenile idiopathic arthritis (JIA); the impact of JIA on family quality of life; the impact of social support on quality of life; the organization of care and; the transition to adult services.

Section two considers:
The supportive structures underpinning the study and research methodological issues.

Section three examines:
A proposed model of care for young people with JIA.
6.2 Section One

6.2.1 The Impact of JIA on the Bio-psychosocial Well-being and Quality of Life on a Young Person with Juvenile Idiopathic Arthritis (JIA)

The quality of life of young people with the chronic disease of juvenile idiopathic arthritis in this small scale exploratory study compared positively with a peer group without JIA. The questions asked of the young people related to the here and now, and also several other timeframes within the previous 12 months. The young people related that they were happy and could do all the things they wanted to do, including meeting and having fun with their friends. They wanted to be the same as their friends and treated the same way as their peers. This is consistent with findings from other research with young people (Blum et al. 1991; La Greca 1992; Shah 2001; Rechner 1990; Weekes & Kagan 1994; Woodgate, 2005).

Acknowledging the difficulties but despite having difficulties, parents too considered that globally their children had a good quality of life. This included not only their physical and health related quality of life but also the social dimensions and social realities of the lives of their young people. Clinicians acknowledged the complex health issues that many young people with JIA had, however from the perspective of many years in clinical practice they were of the view that the health outcomes, functional status and quality of life of many young people with JIA had improved significantly during the course of their own clinical practice life-time and were projected to improve into the future as a direct response of the availability of newer medications.

Juvenile idiopathic arthritis, like many chronic diseases, can require ongoing management over a period of years or decades (WHO 2005). The illness was unpredictable for many young people who participated in the study, with the consequence that the disease trajectory was not always linear. Physical function, global health assessment and fatigue are important constituents of rheumatology assessment (Khanna et al. 2011). Monitoring for early signs of a flare-up became routine in order to prevent or suppress, if possible, a ‘full-
blown' flare-up. Rapid escalation of symptoms required rapid mobilisation of resources to suppress the flare-up. This required ongoing surveillance and a high level of vigilance by family members, usually mothers, to prevent or minimise such an event or reduce the possibility of complications of a flare-up. In this current juvenile arthritis study one parent spoke of recovery time from an acute flare-up being protracted, taking 6-12 months to return to the level of energy prior to the onset of the episode. This had significant implications for the young person and their family including absences or reduced number of hours at school. Loss of school hours would have to be made up and these would impact on leisure time activities. All of the parents in this study were working consequently time away from the work-place caring for an ill-child or attending a health appointment had financial implications due to loss of earnings.

Living with juvenile arthritis meant living with the challenges of fluctuations of wellness and illness. The acuity of the illness, the frequency of the flare ups and the response to treatment had implications not only for the quality of life of the young person with the illness but also for family quality of life. This is congruent with findings from Donnelly (1993) and Patterson (2001) who refer to the ever-changing continuum of living with a chronic disease and its inherent instability, creating within the person with the disease shifting perspectives of wellness, periods of illness and periods of acute exacerbations of illness. The dynamics of the disease can have consequences for family members with and without the illness, causing uncertainty in the home environment for family members due to the loss of the normal routines of family life. The dynamics of family situations can become more complicated as some children try to hide their pain and distress in an effort to cope and to avoid being a burden to the family (Altschuler 1997).

These fluctuating perspectives can also be impacted upon by other variables including the disposition of the person and their response to the situation. For
example, an individual response may be to take control of the situation (Barroso 1995:144) and shift ‘from a victim of circumstances to a creator of circumstances’, thus keeping the wellness perspective or degrees of the wellness perspective to the foreground of their life and the illness perspective or degrees of the illness perspective to the background of their life. This strategy was noted to be used from an early age by some of the young people in the study. Scheier et al. (1986) suggest that this adaptive problem-focused coping strategy is associated with the disposition of optimism. Nes et al. (2005), discussing this concept, suggest that optimists may persist and try harder to attain their goals despite adverse life events and other obstacles along their goal pathway.

Many parents spoke of the difficulties their young people had in disclosing their arthritis to their friends. Juvenile idiopathic arthritis is frequently invisible or not immediately obvious to those who are not specialists in the discipline of rheumatology, consequently the young people had options whether to disclose to others that they had the condition. There was also the possibility that they treated it like a temporary ‘interruption’ in their lives rather than a chronic disease (Charmaz 1997). Concerns relating to being treated differently by their friends were strong considerations in their ambivalence to disclose. Patterson (2003:461) states ‘enormous effort is invested in appearing “normal” despite the conflict that this invokes for the person.’ This could have the consequence of ‘suffering in silence’ in a public situation. The fact that juvenile arthritis is frequently an invisible and silent disease also has implications for young people’s reluctance to discuss their health issues with others whom they perceive may not understand the situation. While the young people stated that they wanted to be treated the same way as their peers, their parents revealed a more complex scenario. Low stamina levels and fatigue, for example, were issues that parents were aware of but did not necessarily want to draw the young people’s attention to. Instead, parents would engineer to normalise the young people’s lives by anticipation of their needs, for example, pacing the young person’s day by reducing the number of hours at school, or reducing the
amount of walking the young people had to do by giving them lifts and dropping them off to the nearest point of entry to buildings. All of the parents and the clinicians spoke of the importance of the young people mixing and meeting with their peers and being as socially active as possible.

Parents made the point that when their children were ill they were frequently at home and interaction with peers was more limited. On return to school their condition had improved and they were able to resume their ‘public identity’ of appearing ‘normal’ (Strauss 1975). Meier et al., (2000) suggests that teenagers with a chronic disease often try to keep up appearances and behave in a socially desirable way in an effort to appear ‘normal’. Other writers have commented also on the impact of deviations from the normal, such as chronic disease, and the potential for associated health-related stigma (Goffman 1966; Bury 1982; Charmaz 1983; Bury 1990). The interviews with parents revealed a more nuanced picture of their children’s lives. If there are disparities between parental and children’s perspectives there is a possibility that the parents are revealing aspects of both their children’s ‘public’ and ‘private’ identity and their children are revealing their ‘public’ identity only in an effort to appear ‘normal’. Normalizing the illness can foster adaptation, it can also have the effect of minimizing ‘identity spread’ and potentially reducing the possibility of stigma associated with the illness (Strauss et al., 1984:81). Scambler (2009:1) states that ‘stigma is typically a social process, experienced or anticipated, characterised by exclusion, rejection, blame or devaluation that results from experience, perception or reasonable anticipation of an adverse social judgement about a person or group’. This can lead to prejudice and discrimination. The stigma can be felt/perceived or enacted. Carter et al, (2011) suggest that stigma is related to social unacceptability, and negative stereotypes. Goffman (1963) refers to this as a situation in which the person’s identity has been ‘spoiled’. Interestingly, in this current study there is evidence to suggest that the young people were aware of their public identity and were trying to protect it by normalising their lives as much as possible sometimes at a physical cost to themselves. It could be suggested that their global quality of life
was given priority over their physical health in some situations. Kelly (1992:391) comments on the difficulties of personal identity and the dichotomy of the ‘tension between the private self and public identity’ and managing to live with a chronic illness. Parents suggested that their children demonstrated maturity beyond their years compared to other siblings in the family. It is possible that young people with juvenile arthritis in this study developed at an earlier age than their peers to differentiate between their ‘public identity’ and their ‘private self identity’. Östlie et al., (2009:666) refers to the ‘struggle and adjustment to an insecure everyday life and unpredictable life course’ of living with JIA which provide complex challenges for the young people to adapt to and cope with on an ongoing basis. They require also that health care providers appreciate these complexities and challenges in order to assist and support the young people to manage and adjust to their circumstances (Östle et al., 2009). Jenkins et al., (2001:8), discussing factors that promote positive mental health development, define it succinctly: ‘a positive sense of well-being, a belief in our own worth and the dignity and worth of others, the ability to think, perceive and interpret, to manage life, to communicate, initiate, develop and sustain mutually satisfying personal relationships’. The importance of positive mental health is also encapsulated in the adage ‘there is no health without mental health’. Parents provided many examples of their young people endeavouring to maintain their psycho-social well-being and promoting their mental health despite numerous challenges.

Milz’s (1992) concept of ‘healthy ill people’ advocates the development of life skills and strengthening personal cognitive coping resources, such as optimism and a sense of humour, in order to optimise their life opportunities. An informal observation of the researcher of the young people who were participants in the study was that they were very co-operative and articulate. The questions asked by them of the researcher during formal and informal interactions were searching and direct, requiring an open, honest and direct response.
During the interviews with the parents of the young people with JIA, frequently and without prompting, they compared traits and attributes of their child with arthritis with other children in the family. Interestingly, it was not clear from the survey questions and results in this study, of whom did the young people with arthritis use as a frame of reference or as a standard of comparison. This is significant because there is no clear baseline, no previous point of reference for comparison (Rapkin & Schwartz 2004). It is possible that they compared themselves with their prior functioning. Eiser and Morse (2001) reviewing the literature relating to inter-rater reliability of parents rating their child’s health-related quality of life identified a number of methodological issues including the specific measures used and the domain being measured, other issues included the location of completion of questionnaire and lack of parallel content in the instruments being used by parent and child can contribute to inconsistent perspectives. Sawyer et al (2004) investigating the relationship between health-related quality of life, pain and coping strategies in JIA concluded also that parent and children have differing views of perceived quality of life with parents reporting significantly lower scores than children on five of the eight scales  Upton et al., (2008:910) suggest that parents and children ‘contribute different but valid information’. Parsons et al., (1999) suggest that the debate is not about reliability and validity or who is right or wrong but how both of these perspectives contribute to the understanding of paediatric health related quality of life. The researcher needed to be clear of the aim and rationale of including these perspectives in the study. In this current study of juvenile arthritis the aim was to gain a richer understanding of the quality of life of young people with arthritis through the inclusion of different perspectives. It is acknowledged that there will be parental variance in aspects such as awareness, sensitivity and tolerance in their perspectives (Upton 2008).

Ward et al., (2011) suggest that the parents of young people with JIA reported sleep disturbances in their children, however in this current study of juvenile arthritis one young person only reported sleep disturbances, however it is possible that they had limited awareness of these disturbances. Assessing
chronic pain levels can be problematic in young people with chronic diseases due to many factors including the habituation of pain behaviours, reduced or increased response to painful stimuli (Von Baeyer & Spagrud 2007; Lootens & Rapoff 2011). In the current juvenile arthritis study direct and indirect questions relating to pain were asked to ascertain how significant the pain was, for example, ‘Does pain stop you from doing what you want?’ and how it impacted on the young person’s quality of life, ‘Are you able to run and move as you like?’ Cognitive components such as comprehension, frame of reference, standards of comparison, reporting and response selection are significant factors in quality of life appraisal (Tourangeau et al. 2000; Rapkin & Schwartz 2004). Unlike the young people, their parents reported the visual cues of pain and indicators of the disease, including stiffness, avoidance of the use of limbs, not moving and remaining still, fatigue and low stamina levels. The parents in this study indicated their points of reference and their comparator groups when they made comparisons between ‘good days’ that the young person had and ‘bad days’.

6.2.2 The Impact of JIA on Family Quality of Life

Juvenile arthritis can impact on families on many levels including physically, emotionally, financially and interpersonally (Packham 2004). Families are expected to cope, often with little formal support. The young people and their families in this current juvenile arthritis study were encouraged to self-manage their treatment regimen as early as possible after diagnosis. However, several young people and their families in this current juvenile arthritis study were encountering difficulties with their medication regimens. They may not have known how to seek help from other sources such as the General Practitioner (GP) or practice nurse or did not feel able to do this, perhaps due to financial or other logistical constraints. Whatever the reason, they as families were incurring severe stressors due to medication regimens. Ongoing follow-up to assess how families are managing medication regimens needs to be integrated into the care and management of these young people and their families by health providers to minimize anxiety and stress and to strengthen coping strategies.
The normal demands of life can create stresses and strains on families; however, superimposing the illness of a family member on this situation can impact on a family’s coping capacity (Cohen 1999). Charmaz (1981; 2010) and Bury (1982) refer to the interruption, disruption and intrusion of chronic disease. This interruption and disruption certainly occurred in families of a young person with juvenile arthritis. Jerrett’s (1994) study focused on the perspectives of the parents caring for a child with JIA. Initially following diagnosis, there was confusion and turmoil and a struggle to understand their new reality and their new role, followed by learning to manage this new reality and then taking charge of the situation. Monitoring the young person watching for the cues and reading the symptoms of the chronic disease were reported by Timmermanns and Friedin (2007) in their study examining young people with asthma and their carers similar to the findings in this current study. Parents had to shift their perspectives from caring for a well child to caring for a child with a chronic disease, Jerret (1994:1053) stated ‘The child’s therapy became an indelible and inescapable part of the parents’ reality’. This study published in North America, Shaw’s work during the 1990s, and Britton’s work in the early 2000’s reflect many of the findings of this section of this current juvenile arthritis study relating to the information needs of parents particularly pre-diagnosis information needs. This would suggest that evidence is available, however there remains a dis-connect between the information being available and ameliorating parental concerns through improved communication channels. In the current study of juvenile arthritis there was also a sense of grief and sadness in the family circle when another episode of arthritis occurred. This was related to the loss of normality for the young person and their family life. It was also associated with the pain and suffering, loss of energy and fatigue that the young person was enduring. During an acute ‘flare-up’ there was a sense of being ‘immersed’ in the illness (Charmaz 1991). The families gave the sense that coping with the symptoms of the disease and managing it could be all-consuming and energy depleting. The literature reflected the coping strategies associated with living with JIA of positivity and being proactive. Interestingly, there was less evidence in the literature of reflecting the grief and sadness of the
family circle when another episode of this chronic illness occurred (Charmaz 1991, Shaw 2006).

One parent summed it up succinctly when asked how family life had been impacted by the diagnosis of their child with a chronic disease. ‘Everything has changed’ was the reflection. The fluctuating nature of the disease can create uncertainty for all family members. It can also create a spectrum of responses from parents. At one end of the scale there can be ‘over-protection’, and at the other end of the scale there can be emotional withdrawal from the situation due to ongoing family tensions with the young person, often related to disease control measures (WHO 2007). Parents in this study endeavoured to get a balance between protecting their young people and promoting their sense of autonomy. The reaction of parents to their situation also ranged from concern, anxiety and guilt to frustration and anger relating to care and service provision. Similar findings have emerged in other studies (Lee & Rotheram-Borus 2002; Kurnat & Moore 1999; Chernoff et al. 1999). Brown (1998) suggests often young people can not always articulate their concerns and their parents can provide a crucial contextual perspective of their child’s situation, however Drowe, (2001) suggests that this ‘discursive othering’ can be problematic if the young person’s views are not included in the discussion. Children may not be able to articulate their concerns but can be affected by their parents’ distress. Altschuler (1997:85), discussing the issues and the complexity of communicating with children relating to illness, states that ‘children are incredibly good information processors. Shifts in emotional tone in the home are noted: where no information is provided, they evolve their own stories which are probably more frightening than reality’. Evans et al., (1992), acknowledging that each family is unique, suggest that greater amounts of information provided at a cognitive-appropriate level can mitigate against higher distress levels in young people with the illness and their siblings.

Adaptation was central to living with juvenile arthritis in this study. Coping strategies are associated with adaptation to a situation (Lazazrus & Folkman
1984). Some of the adaptations were major, others less so; some were physical, others cognitive, but they could all assume some significance in a family setting. All members of the family were implicated. Some examples included adapting lifestyles to medical regimens, managing the condition, and organizing the pace of the day for the young person with JIA. Family life events such as day outings, holidays and home routines were planned around the illness of the young person. Spontaneous life was a rarity. Family life needed to be deliberate, planned, conscious and frequently constrained. During a flare-up it took the young person longer to do everything. This could give rise to frustration and upset as their time perspectives had changed and temporal incongruence had developed (Charmaz 1997). Apparently small but significant adaptations to reduce frustration and upset included adapting clothes to avoid buttons and laces, adapting furniture (bed, chair, desk), and increased heating in the home were reported by parents in this study. Parents in this current juvenile arthritis study reported that warmer micro-environmental climates were associated with less pain and stiffness. A strong finding in this study was that significant consideration, accommodation and adaptations were made by families to reduce the impact of the disease on the young person with JIA.

Illness can create a sense of loneliness (Altschuler 1997). An example of promoting inclusion and avoiding isolation in this current juvenile arthritis study was giving the young person with arthritis priority over the couch or day-bed in the living room. This enabled the young person to watch television with other members of the family, which kept them connected and avoided isolation. It also promoted stimulation and interactivity with the family. Families were creative and proactive in their problem-solving adaptations and improvisations. Parents also spoke of teachers quietly and sensitively accommodating their young people without drawing undue attention to the young people in an effort to promote their inclusion, socialisation and integration and also helping the young people to meet their academic targets. Promoting a sense of connectedness with the school and a positive school experience can assist with academic performance and can contribute to long-term life opportunities.
(Mentality 2003). Parents and teachers were creative and their solutions were infinite in order to be inclusive, maintain the young person's socialisation, and avoid isolation.

In this study, parents in general considered that their children had a good quality of life. This could be related to improved disease-suppressive medication reducing the impact of the disease. The perspectives of parents in the study provided a longitudinal overview of the young person's illness and its impact. Parents were able to assess the impact of the condition on the young person's quality of life. They were also able to compare the quality of life of the young person with JIA with that of other siblings. A longer life experience also provided parents with the capacity to consider the long-term implications and the impact of juvenile arthritis on the young person. These are perhaps some reasons why parents in the literature often report significantly lower health-related quality of life than their young people with a chronic condition (Sawyer 2005). However, as Sawyer (2005) suggests, it is possible that young people with arthritis have adapted better to their illness than their parents have reported or perhaps the young people could be internalizing their problems and not reporting them. Another possibility is that the right questions are not being asked of the young people.

Clinicians in this current juvenile arthritis study reported that in the recent past (up to 20 years ago) they would have had many patients with JIA who were wheelchair users, but this is no longer the case. Chronic pain and joint stiffness is a significant feature of juvenile arthritis, however, newer studies have noted reduced pain levels with the use of newer biologic medications with a resultant improvement in quality of life (Pincus et al. 2008). Parents were aware that the medications were improving the young people’s quality of life. They understood that the medication was not curative; it was suppressing the disease, and should a young person have a flare-up the return time to full recovery could be lengthy. Parents and clinicians recognised that rehabilitation time and treatment are improved if the appropriate allied resources are available. In the long term it
makes social and economic sense to provide appropriate services to effect an improved recovery time, as this will enable young people to promote their life opportunities, allowing them to be more productive and to obtain gainful employment in the future.

6.2.3 The Impact of Social Support on Quality of Life

Cobb’s (1976:310) benchmark work of many years ago resonated with the researcher when the young people and parents spoke of the significance of direct and indirect social support:

‘We have seen strong and often quite hard evidence repeated over a variety of transitions in the life cycle from birth to death, that social support is protective. The very great diversity of studies in terms of criteria of support, nature of sample and method of data collection is further convincing that we are dealing with a common phenomenon.’

Social support, which is strongly linked to quality of life, was important for the young people with JIA - particularly social support and acceptance from their peers. There are many forms of social support. Cohen & Willis (1985) suggest that emotional support relates to empathy, care, love, trust, understanding and listening. Instrumental support is concerned with tangible assistance and services, for example child care or financial assistance. Informational support consists of advice and information, improving knowledge and understanding and consequently assisting with coping with adverse events. Social support is provided by members of the social network. All of these forms of social support were found during this study when considering the micro-perspective.

In some situations not all members of the social network provided positive support (Rose, 1997; Åstedt-Kurki et al. 1999). Parents and young people alluded to this, suggesting that not everybody understood their situation. This is consistent with the literature which suggests that as JIA is relatively
uncommon, parents can feel emotionally and socially isolated due to an under-appreciation of their particular situation (Barlow et al. 1998; Hagglund et al. 1996). Some commentators suggest that a positive family environment can be a critical factor influencing positive well-being and promoting coping with chronic disease (La Greca et al. 1995; Price 1996; Kyngäs 2000).

Kessler and McLeod (1985), acknowledging the complexity of relationships, concluded in their review of the literature that social support provides a buffer effect, particularly in chronic illness situations. This buffer effect has positive implications for quality of life. It is possible that parents, in their role of advocates, provided a social buffer effect for their children by reducing or avoiding potentially difficult situations for them, or by being proactive in anticipating difficult interactions and creatively circumnavigating these situations to maintain their children’s socialisation and inclusion. This was done by being proactive and by anticipating or reacting to their physical, social and emotional needs. However, this often required extra effort, extra energy and extra creativity by parents. The young people were in a period of transition of maturing into adults and developing their own sense of identity; consequently it was hard for parents ‘to get it right’ all of the time, and at times parents’ efforts were under-appreciated. One striking example of dedication, love and ingenuity was of a mother with a young person with JIA at college abroad who did not have the required medication and who phoned home. The mother got the medication, got a flight to the nearest city, delivered it to her child and returned home that day. Interestingly, Charmaz (1994) and Williams (2000) noted in their studies gendered patterns of behaviour of mothers to sons which were different to their patterns of behaviour to their daughters. Transition into taking full-responsibility for self-management was an evolving process, parental boundaries evolve and relationships evolve as the young people mature (Stinson 2008). However, it is possible that the young people did not appreciate how much their parents had buffered them.
Bø (1996) and Cottrell (2007) suggest that adolescents have a group of stable relationships, frequently comprised of parents, best friends, peers and teachers. This group provides the young person with a secure protective social support base which can assist with social and emotional adjustment during adolescence (Coates 1987; Furman and Buhrmester 1992; Levitt et al. 1993; Urberg et al. 1995; Levitt et al. 2005; Milevsky 2005). Cobb (1976) asserts that these relationships can assist in communicating to the young people that they belong and are valued, cared for and loved. During a period of change and development, or adverse circumstances such as an acute episode of illness, these relationships can provide a sense of stability for the young person as other circumstances shift or unsettle their equilibrium. These protective and supportive systems can assist with the development of resilience (Luthar et al. 2007; McDermott and Graham 2005; Lehtinen et al. 2005; Everall et al. 2006; Drapeau et al. 2007; Bottrell 2009a; Bottrell 2009b).

Gilligan (2000:37) states that ‘a resilient child is one who bounces back having endured adversity, who continues to function reasonably well despite continued exposure to risk’. Gilligan (2005:38) further suggests that if there is an accumulation of adversities, the young person will ‘buckle under the strain’. He stresses the importance of reducing the number of problems that the young person has and that ‘this may have a disproportionate and decisive impact’, as even small positive experiences may be a positive ‘turning point’ (Clausen 1995) for the young person. This has significance for healthcare providers who are treating young people with the chronic disease of juvenile arthritis, as a person-centred global approach to their care is required to protect and promote their health. Changing treatments or planning care provision needs to be viewed in the context of the young person’s current life situation and competing demands on their time, for example changing treatments prior to school examination periods. Encouraging young people to build skills and competencies, encouraging confidence, capacity and supportive environments are important factors in the development of young people. It is apparent from this study that when caring for young people with the chronic disease of juvenile idiopathic
arthritis the approach to service provision needs to be holistic and person-centred.

Positive nurturing is important for all children but takes on an added significance when the young person is coping with adverse circumstances such as juvenile arthritis. Gilligan (2005:45) suggests that seemingly small gestures can make a significant difference. ‘The rituals, the smiles, the interest in little things, the daily routines...all of these little things may foster in a child the vital senses of belonging, of mattering, of counting.’ This positive nurturing needs to come from all of those in contact with the young person whether in the home, school, community or healthcare setting. There is a need for those working with young people with JIA, for example in a healthcare setting or an education setting, to understand their illness and their particular situation, to have good communication skills and be able to connect with the young people to appreciate their concerns in order to optimise each individual’s health and well-being.

6.2.4 The Organization of Care

Healthcare provision was viewed by study participants from a micro-perspective, a meso-perspective and a macro-perspective. Parents discussed their role as their child’s healthcare manager. This role could include actively liaising with several health professionals, co-ordinating services, arranging assessments, maintaining files and information, and constantly updating all the interested parties. Parents provided a wealth of information on the nuances of the disease, for example the triggers and the learnt best practice of daily care for their young person with arthritis. At times the parents found their role of healthcare manager overwhelming.

The parents’ principal concern was their children’s welfare. The role of advocate was viewed as a necessity, but parents often found it a difficult and frustrating role when trying to access services for their child. This was found to be particularly difficult when also endeavouring to care for a child during an
acute exacerbation of the illness. Parents often perceived themselves as very much alone at these times. Parents commented that their route through the healthcare system was less than smooth but was instead fraught with inconsistencies and difficulties.

Accessing resources, either informational or physical, was an area that parents found problematic, particularly in the early days of the illness, with resultant stressors for the families. Parents in this juvenile arthritis study were looking for more explicit information on the daily care of their child particularly when their child was first diagnosed with the condition and they were inexperienced and lacked detailed knowledge of the provision of care. There was not enough detailed information on how to care for their children in an acute flare-up situation; giving medicines on a daily basis to a reluctant or refusing child; giving medicines to a nauseated child; communicating effectively with healthcare professionals; and accessing part-time teaching for a child who had missed a lot of school. Guidance, if any, was not comprehensive. Accessing information and advocating for resources from health service administrators were two adaptive strategies that parents, by necessity, utilised; however, the route was seldom smooth and frequently circuitous (Ombudsman for Children 2008). Effective and efficient information systems offer improved potential for more integrated and co-ordinated service provision for young people with juvenile arthritis, for example telemedicine. Strengthening the collaborative and partnership processes between families and healthcare providers could be supportive in assisting families cope with the daily stressors associated with juvenile arthritis; for example, discussing the cues of a ‘flare-up’ with a specialist provider could reduce the acuity or complications of a ‘flare-up’ and could also ease the responsibility of care for the parents.

Parents were of the view that they were not in control of the situation during the early days of their child’s illness prior to diagnosis and during other times of acute illness. Horn, Feldman & Ploof (1995) suggest that a sense of control of
the situation is critical for parents, if their capacity to cope is to be strengthened. Not only were they not in control of the situation, their engagement with healthcare service providers prior to a secure diagnosis of JIA was less than reassuring.

The Ombudsman for Children (2008) has also referred to inconsistencies in service provision and difficulties for children with chronic illnesses. Wortman & Conway (1985) suggest that socially competent people may be more effective and have greater capacity to access resources, for example negotiating the health system and consequently obtaining better care and treatment. This gives rise to the potential issue of inequalities in the provision of health care and the concern that if parents are not skilled negotiators and competent advocates there are potential adverse implications for their children. In the United States, in response to these issues, a new role of healthcare advocate has emerged (HCA). Vasserman-Stokes and Cronan (2012:27) define HCAs as professionals ‘who represent the interest of the patient and work with them to reduce the complexities implicit in the treatment of chronic illness’. It is possible that such a role will emerge in this country to assist families in navigating the healthcare system.

During the interviews with consultants and specialist nurses, clinicians explained how difficult it could be to diagnose juvenile idiopathic arthritis. They referred to the large number of other diseases that needed to be excluded prior to a secure diagnosis of juvenile arthritis being confirmed. At a time of stress, anxiety and confusion for parents and their children, this message did not seem to be clear to parents. The time prior to securing a diagnosis was a time when anxious families were seeking clear communication relating to the pathway of diagnosis, for example an algorithm that sets out a diagnostic pathway. Parents needed someone to demonstrate that they were taking control of the situation. They did not always perceive this to be the case. Parents also needed to be able to understand the diagnostic plan pathway for their child. Parents found this area of communication to be problematic and in need of further consideration.
by clinicians. If parents were dissatisfied with their physician-patient relationship they changed doctors. None of the parents indicated that they confronted the physicians with their concerns. Post-diagnosis of their child with juvenile arthritis, parents recognised in themselves that they too were also coming to terms with the limited medical knowledge relating to the disease, the limited treatment regimens and options, the chronic nature of the disease itself, and the fact that there was no ‘quick fix’ solution.

Michaud et al., (2007) in the WHO report ‘The Adolescent with a Chronic Condition’ discusses the importance of youth-friendly healthcare environments and the need to have ongoing trusting relationships with healthcare providers, as these can support adherence to regimens and also assist the young person to take responsibility for the management of their own situation as they progress into adulthood.

Having been diagnosed with arthritis and referred to a specialist rheumatology clinic, the young people in the study considered that they were listened to by their consultants and that they were accorded appropriate respect at consultations. In some cases the young people did not get the opportunity to speak with the doctor or healthcare providers alone as a matter of routine. This is an area where there is potential to develop the communication skills of young people and give them an opportunity to speak directly to the clinicians. However, it is recognised that this is not always feasible depending on the young person and the circumstances. Clinicians also need to be aware of the power asymmetry between young people and themselves and the issues that adolescents may have in articulating their concerns (Kieffer 1984). Direct access to consultants (either by phone or email) was considered a ‘lifeline’ and viewed as an act of great trust, and was deeply appreciated by families. The newer telemedicine and tele-monitoring systems offer patients the potential in the future of adjunct consultations with specialists (New Zealand Medical Association 2008).
Parents and consultants spoke of the specialist nurses very positively. The collaborative approach of care providers and the accessibility of specialist nurses were found to be very helpful for parents and young people. For example, queries that were considered insufficiently significant to contact the consultant about could be dealt with by the nurses. The use of telephone help-lines and texting were two modes of communication that were found to be particularly useful. Ongoing advances in electronic communication systems will doubtless provide new resources and modes of communication and means of assessing the young people’s quality of life, physical function and global health assessment including levels of fatigue. It will be important that healthcare providers have access to these information and media systems to improve the care of young people with arthritis (O’Neill 1998; Forsstrom & Rigby 2000). Other advances in medical devices and medical technology which are less invasive on the person could be of greater assistance in the future in diagnosing, clinically assessing and monitoring disease activity in the young person with JIA and could also be assistive in contributing to decisions to taper medication (McKay et al 2010). Rebollo-Polo et al. (2011) suggest that imaging modalities are clinically assistive in joint assessments. Doppler ultrasonography in the Rebollo-Polo (2011) study demonstrated ongoing pathology in young people with JIA who met the clinical criteria for remission. Rebollo-Polo et al. indicate that this finding of pathology is suggestive of persistent inflammation. This finding if replicated in other studies and found to be persistent inflammation due to disease activity has important implications for the care and management into the future of young people with JIA. This finding also indicates the importance of listening to the patient as some studies have suggested that the degree of pain experienced by young people does not always correspond with the degree of inflammation indicated by clinical markers (Weiss et al. 2005).

Healthcare record keeping and informatics were areas of concern for parents; lack of sharing of data between clinicians and lag times in reporting of data were sources of frustration. Parents and clinicians spoke of the fragmented data, the complicated processes of the system and the duplication of data.
recording. These all had an impact on clinical practice and clinical effectiveness, as well as the patient ‘experience’ of the health system. Parents found these forms of organizational difficulties a source of high frustration, particularly when their child was coping with an acute flare-up of arthritis. Parents considered that life could have been made easier for their children and themselves if the health system had been more focused to their needs. At a time when their energy needed to be directed towards their ill child it had to be channelled elsewhere to access services or medications. Sometimes parents only had sufficient energy to give to their child and consequently they disengaged from communicating with healthcare providers until their child had recovered sufficiently, and they too had recovered their energy. Parents did not consider that the care system was built around their child’s needs; rather the provision of care was fragmented, complicated and uncoordinated.

McWhinney (1989) refers to the importance of healthcare providers entering their patients’ world and endeavouring to consider life from their patients’ perspective. One example relates to competence in taking blood (venepuncture). Young people with JIA, particularly those on the newer biologic medications, need to have their blood levels checked regularly, particularly for analysis of liver function as the liver can be affected by these medications. Recommendations for the use of these disease modifying anti-rheumatic drugs and biologic agents are regularly reviewed these include indications for use monitoring of side effects and assessment of clinical response (Singh et al. 2012). Parents spoke of the under-appreciation by clinicians of the impact of anticipatory distress, invasive procedural distress and the side-effects of medications (Claar, Walker & Smith 2002). Parents were of the view that novice learners in phlebotomy should not be allowed to do invasive procedures such as taking blood (venepuncture), as the consequences of a poor experience for their children were frequently long-lasting. Another example is the colour yellow. This is a colour that many young people on the oral medication Methatrexate do not like and associate with gastrointestinal upset and nausea. The tablet is yellow in colour. One parent in this current juvenile arthritis study whose child
had been on oral Methatrexate nearly ten years ago reported that the young person still had difficulties with anything coloured yellow. It was referred to by both parents and clinicians and is a known phenomenon in the literature. Medications or routes of medication may have been changed by clinicians but the young people remained sensitised to the colour yellow. In this study desensitising the young people was recognised as an issue by clinicians but not a priority concern. Further research may need to be done on this issue to explore if it has been under-reported in the Irish context due to lack of allied health services, including psychologists. However, as Haverman et al (2012) and Seid (2012) indicate, the health related quality of life of young people with JIA can be affected not only by disease activity but also by medical interventions. Lovell et al. (2011) discuss the importance of appropriate services for young people with JIA in the United States of America, they discuss also the significance of the quality of the services available to these young people. They suggest the need for a number of components including a JIA registry, ongoing uniform assessment, longitudinal data collection, the development of a network of paediatric rheumatology centres to incorporate proposed quality measures related to disease control, safety monitoring, assessment of self-efficacy, access to services and patient/parent satisfaction. In Ireland, baseline data and serial acquisition of data and collaboration of centres could be assistive in improving the disease status and quality of life of young people with JIA.

**6.2.5 Transition to Adult Services**

The importance of a planned transition from paediatric to adult services was referred to by parents. The use of integrated clinical pathways, clinical models of care and interdisciplinary co-ordination could assist in the provision of a more holistic patient healthcare pathway with improved patient outcomes (Ellis 2010). Improved continuity of care is affected by good protocols, good care co-ordination and good information, for example patient registries (NHPAC 2006). Patient-centred support is needed to assist people manage their treatment regimens but it is also needed to support people living with a chronic illness (Davidson & Cruikshank 2009). The young people need to be educated and
supported to manage this new role (Elwyn et al 2003; Thon & Ullrich 2008). Hersh et al. (2009:7) highlight the importance of a 'seamless transfer of care' of young people transferring from paediatric services to adult rheumatology services and indicates that flare-ups of the disease are common at this time.

Doctor patient relationships have changed significantly during the past 20 years and this is of importance especially as young people transit from paediatric services to adult rheumatology services. Increasingly physicians actively explore patient views and expectations and acknowledge the expertise of patients and knowledge of their disease (Stinson et al. 2008).

The literature suggests that transition is a process and that all parties need to be prepared and ready to make a transit from one setting to another setting (Blum, Garell & Hodgman 1993; Shaw et al. 2004; Shaw et al. 2005; McDonagh et al. 2007; Stinson et al. 2008; van Staa 2010). This requires the young person to have a ‘developmental readiness’ (Viner 1999; Betz, Redcay & Tan 2003; McPherson, Thaniel & Minniti 2009). The American Society for Adolescent Medicine (Blum et al 1993:570) states the process of transition is ‘the purposeful, planned movement of adolescents and young adults with chronic physical and medical conditions from child-centred to adult-oriented health care systems’. Mc Donagh (2008) highlights the importance of focusing on the young person first and their arthritis second. The timing of transition from paediatric to adult services is often a time of other changes in the young person’s life, perhaps changing from a home location, perhaps a new job or going to college some distance away often placing significant challenges on coping and adaptation strategies (Östlie et al. 2009). Compliance with medical regimens had been overseen by parents during their earlier years. Adherence to treatment regimens can be problematic during this transition time (Pai & Ostendorf 2011). There is a strong risk of the young people dropping out of the healthcare system during this time (McGill 2002; Hazel et al. 2010). This has been noted in other populations transferring from paediatric to adult services, for example cardiac populations (Reid et al. 2004) and diabetic populations (Van 2008). Successful transition can be affected by many issues including
issues relating to the patient, the health system organization, paediatric, rheumatologist and team, the adult rheumatologist (Fox 2002; Tucker & Cabral 2005, BSPAR 2009). Preventing harm and promoting the health of young people with JIA are central to transition clinics. One important issue which may not seem central to the lives of young people in their late teens early twenties with JIA has been selected as an example of the importance of ongoing monitoring of young people with JIA into adulthood, the example is low bone density. Low bone density may seem to be peripheral to the lives of young people because concerns relating to it are not immediately obvious nor may they appear to be impending. There are many factors that can influence skeletal peak bone mass including chronic diseases such as juvenile arthritis, in particular disease activity and the duration of the disease. Skeletal maturation normally occurs during the second decade of life, if the foundation of peak bone mass is affected it can increase the risk of bone fracture and osteopenia in later years (Lien et al 2003). Preventing harm into the future is part of the work of rheumatology clinics; however, the need for ongoing assessment and monitoring may need to be ‘packaged’ more attractively to encourage understanding and participation by the young people. In rheumatology, young men and those with less severe JIA were at greater risk of unsuccessful transfer, suggesting the need to target young people in these categories (Hazel 2010). A predominant need to appear normal and intentional avoidance of activities that could be an indicator, identity marker or provide confirmation of difference are thought to be contributory factors to this situation of avoidance of being perceived negatively or associated with a marker with negative connotations (Knafl & Deatrick 1986; Ullrich et al. 2003; Charmaz 2010). Some of the reasons for disconnecting from the health services may be intentional others may be unintentional including taking responsibility for health care is low on the young people’s priority list as parents had assumed responsibility for these matters since their childhood and there is an expectation that this will be continued (Tucker & Cabral 2005). There is a need for further study in this area to explore this situation and issues such as communication skills and engaging with young people, problem solving skills, barriers to adherence to treatment regimen
including delayed beneficial effects and negative side-effects (Rapoff & Lindsly 2007; Stinson et al 2008). It is possible that low visibility of the disease or lengthy periods without the disease could reduce the certainty of having the disease further reducing the young people’s concern for adherence to the treatment regimen (Charmaz 1991). It seems also that some elements of transition could occur at an earlier age with parents and others available to monitor and mentor the young people, for example making and confirming routine appointments with health care providers (Stinson et al. 2008).

Parents in this current juvenile arthritis study spoke of the supportive networks that were available near home and the concern that these may become less easy to access due to geographical re-location by the young people. The loss of normal supportive networks and the new, exciting and often overwhelming stressors of a new job or going to college could impact on the young person adhering to treatment regimens. Healthcare providers need to appreciate the complexity of change that can be occurring for the young person at this time. Gilligan (2000) suggests that small measures can be disproportionately significant when working with young people. Good communication skills at the first meeting with healthcare providers in the adult setting could offset one stressor for the young people. A liaison or reference person between the paediatric service and the adult service with direct responsibility for the young person’s smooth transition into the adult service could be helpful in addressing this situation. Marginal alterations do not always effect change. However, it is acknowledged that small changes in a system can often have a ripple effect on the whole system with the potential for a spiral of positive change (Gilligan 2000).

6.3  Section Two

6.3.1  Health Promotion Theoretical Framework Underpinning the Study

At the early stages of this small-scale exploratory study it was supported by Tannahill’s model of health promotion (Downie et al. 1996). This model
includes the concepts of health protection, prevention of ill health and health education and has implications for well-being and quality of life which were the focus of the study. Tannahill’s model provided a structure and context to the study.

As the study advanced, there was greater interaction between the researcher, young people with JIA, their parents and clinicians it became clear that Tannahill’s model was not encapsulating all of the perspectives that were emerging. To complement Tannahill’s model the Innovative Chronic Care framework (ICCF) was chosen as it provided guiding principles on three levels; the micro level, the meso level and the macro level. The micro-level refers to the individual/patients/families and their healthcare providers. It also includes community partners. The meso-level is aligned to the community, healthcare organization including co-ordination of care and the care continuum. The macro-level is concerned with policy and financial issues (WHO 2003; Epping-Jordan et al. 2004; Nolte & McKee 2008). This approach provided the opportunity to consider not only an ‘eyeball to eyeball’ perspective with the young people about their arthritis (micro-perspective) and their healthcare services (meso-perspective), but also gave the study scope to discuss and explore with young people, parents and clinicians the way forward for health services for young people with juvenile arthritis in Ireland, thus gaining a macro-perspective on the situation. This provided the opportunity to explore how treatment and service provision could be optimised (WHO 1988; Bird 1990; WHO 2005). Bronfenbrenner’s (1979) concept of ecology and the impact of multiple physical and cultural influences that shape the development of human beings over time provided significant insights and context to the study. These models positively influenced the study by providing depth and breadth and by illuminating different contexts, perspectives and dimensions.

In terms of policy, the backdrop to the study was Health 21 – the health for all policy framework in European countries (WHO 1998b). In Ireland, the key relevant public health strategies included the National Health Promotion
Strategy 2000-2005 (DOHC 2000) and the National Children’s Strategy ‘Our Children – Their Lives’ (DOHC 2000). These policy documents included a focus on children and young people and had the priority settings of homes, schools, workplaces and communities. All of these priority settings had significance for this study. The ‘Tackling Chronic Health Problems’ strategy document (DOHC 2008) targeted more substantive issues relating to people with chronic diseases in Ireland. This strategy, too, was of assistance in framing the study within a health promotion context.

The Ottawa Charter (WHO 1986) has five pillars for action to promote health: to build healthy public policy, create supportive environments, strengthen community action, develop personal skills and reorient health services. The Charter provided a foundation for this study. Commentators like Antonovsky (1996) were also significant when metaphorically weaving the infrastructure to support the study. Antonovsky (1996) asserts that health promotion has its foundations in a salutogenic view of health. The aim is to increase a person’s health potential at a number of levels: the micro-level of the individual, the meso-level of the community and the macro-level of the entire population. It focuses on coping positively and flexibly with the vicissitudes of life through understanding of circumstances. Antonovsky also posits that people and their environments are intricately linked; consequently there is a need to consider a socio-ecological framework for the approach to healthcare provision (Barry and Jenkins 2007).

6.4 Section Three
6.4.1 Research Methodological Issues
It was recognised from the outset that there were likely to be many challenges to this research project. The number of young people in Ireland with the condition was unknown. Informal discussions with paediatricians suggested that the number was likely to be small but there was no registry to confirm this view. This was the pivotal point to stop any further work in the area or to continue on. The survey to consultant paediatricians and consultant
rheumatologists in Ireland, and one paediatric rheumatologist in Northern Ireland, was designed to provide an estimate of numbers of young people in the country with JIA. This information was important to get an indicator as to whether to proceed with the study. It was known these consultants were very busy and had very little, if any, administrative support in their public clinics. Accessing patient files meant doing a manual search, not an electronic search, therefore requesting very detailed information was likely to reduce the response rate. Despite this the response rate turned out to be high. The survey confirmed the names of the consultants in the country most interested in juvenile arthritis. An unexpected and surprising outcome of the survey was the number of hand-written notes by consultants encouraging and supporting the research study.

The survey provided information that indicated a small number of consultants with relatively high numbers of patients with juvenile arthritis. The results of phase one of the study assisted with the development of the next phase of the study. A decision was made to seek access to these patients through their consultants. Gaining access, approval and support from a consultant and gaining ethical approval for the study were required for each site. Approval from the university research ethics committee was also required. These factors impacted on the start dates at some of the sites as the process could be lengthy. In recent times the ethical approval process has been streamlined; however, there is something to be said for the inquisitorial process of a children’s hospital research ethics committee, whose work is to protect children by asking searching and direct questions. Preparation for these types of situations was of paramount importance.

There is much written about gatekeepers and barriers to research projects. However, once all the paperwork, including Garda clearance, was in order, the main concern was finding the numbers of young people with JIA to meet the study criteria. Parents were very open to participating in the study; so also were young people and clinicians. Everybody that was asked to participate in
the study agreed to do so nobody declined to participate. The consultants at the different sites screened their lists of young people attending public outpatient JIA clinics to identify those who met the study criteria. Private consultation clinics were likely to be more generic and not specifically related to juvenile arthritis. The sample of young people to complete the questionnaire was, however, numerically too small to provide sufficient data to discuss the quality of life of young people with JIA in adequate depth and breadth. There was a significant discrepancy between the anticipated number of young people attending the Consultant out-patient clinics and the number that were actually in attendance. Reflecting on this situation, reasons for this discrepancy include the possibility that the type of arthritis that these young people had was at the milder end of the spectrum and while still on the Consultants lists of patients to be reviewed intermittently most of their care was being delivered at primary level. The level of juvenile arthritis could also be less in Ireland than in other countries as the expected incidence was not aligned with the United Kingdom of 0.1 per 1000 children and a prevalence of 1 in 1000 (Davidson & Cruikshank (2009), or there is a possibility that juvenile idiopathic arthritis which is undoubtedly difficult to diagnose may not have been diagnosed. The Consultants in general provided an open access policy and there was an opportunity to discuss the situation and in most cases to check the records – the sample however remained small.

6.4.1.2 Sample of Parents

The sample of parents interviewed was identified as the parents of a subset of a group of young people with arthritis. The disease status of the young people with arthritis was such that their disease required immuno-suppressant medication and the new biologic medications to suppress the disease. The disease of this group of young people was considered by the consultant to be more moderate than severe, but it was not considered to be mild. The interviews with parents and clinicians provided greater insight and understanding into the lives of the young people. Triangulation of data provided not a complete picture of the situation, but corroboration and
convergence of findings. Triangulation also provided different perspectives, which strengthened the story that was unfolding and emerging through confirmation, comparison and integration of data findings (Johnson & Onwuegbuzie 2004). Hoetker (2005:84) was of the view that ‘the combination of qualitative and quantitative methods allows me to capture a more complete, holistic and contextual portrayal of the phenomenon’ providing complementarity to the understanding of the findings. The perspectives of the interviewees and other sources of data contributed unique angles in the exploration of the health related quality of life of young people with JIA. Combining these data sources assisted in addressing the research question in greater depth, each source contributed information from their particular standpoint, background and area of expertise whether as young people with JIA, parents or clinicians (Shih 1998). A diagram to display the findings (please see Appendix 12) schematically demonstrates the findings from the three groups, young people, parents and clinicians (O’Cathain, Murphy and Nicholl 2007; Creswell and Plano Clark 2007; Fetters et al 2007). The schematic diagram providing a visual anchor assisted in managing, analysing and interpreting the data further (Wendler 2001).

Redfern & Norman (1994:51) list a number of limitations to triangulation including no guarantee of internal and external validity, may compound sources of error, methods selected may not be the right ones, unit of analysis might not apply to all methods, cannot compensate for researcher bias, expensive, no use with the ‘wrong’ question, replication difficult. Many of these limitations relate to other types of research as well as triangulation, for example researcher bias, however using a combinations of methods in this current juvenile arthritis study may be assistive in reducing researcher bias. It was very useful to have Redfern & Norman and other work including Begley (1996) Casey & Murphy (2009) to refer to when considering these issues in order to make the case that the methods chosen for this study were selected specifically to answer this research study question which aimed to explore the health related quality of life of young people with juvenile idiopathic arthritis. To pursue the line of
argument further, following further consideration a subjective decision was made that the advantages of this chosen method outweighed the disadvantages as it allowed for the findings from each source to be complementary and have the impact of strengthening confidence in the results. The purpose of the study was to explore the quality of life issues of young people with JIA in Ireland, a population about which there was very limited information. The intention of the study was to expand the knowledge on the topic consequently the generalizability of the findings was of less significance than testing theories (Wolfe 2008).

6.4.1.2 Reliability of Results
Comparing the parents’ perspectives of their children’s quality of life with their children’s perspectives in this study the two perspectives were generally comparable. This has been found in other small quality of life studies for example Majnemer & Shevell ( 2008) who compared the self-report ratings of quality of life between parents and their children of school age with cerebral palsy. The self-report of the young people and their parents were generally comparable but parental ratings did differ in some domains including school functioning where there was the weakest agreement and the strongest agreement on physical health. In this current juvenile arthritis study the parents were interviewed and did not complete a questionnaire, a numerical comparison is therefore not possible and is a limitation of the study. The key domains of the questionnaire administered to the young people with arthritis were the main subject areas addressed in the interviews with the parents and the clinicians.

Returning to the issue of the sample size, the overall sample size of young people participating in the study was small. This is unquestionably a limitation to the study. Fulton Picot et al., (2001:695) states ‘rare populations are small subsets of the general population – too small in probability samples to allow for multivariate analyses’...‘screening is often necessary to identify a rare population from special lists that constitute the sampling frame’. During the
study efforts were made to increase the sample size by checking and rechecking if the generated outpatient clinic patient lists had changed and if new additions to the lists met the criteria of the study. Informal discussions with consultants suggested that the patients names noted on their outpatient clinic lists were the expected groups for the clinics. A response rate of 60% was a relatively good result however the non-responders the 40% could have been the group with the least good health related quality of life and their views were not captured.

6.4.1.3 Ethical Issues Relating to Children and Research
Grodin and Galntz (1994:5) discuss the concerns relating to children and research. Endeavouring to gain a balance between the vulnerability of children and increasing knowledge about them in an effort to improve their situation can be problematic:

...*presents a powerful tension between two sometimes conflicting social goals: protecting individual children from harm and exploitation, while at the same time increasing our body of knowledge about children in order to develop beneficial medical, psychological, and social interventions.*

The role of parents is to protect and act as advocates for their children; however, there can be power issues relating to young people, parents and collecting data (Morrow & Richards 1996). One reason for using a questionnaire to collect data from the young people was to provide them with the option of completing it at the clinic or taking it home and completing it and returning it by post. Built into this thinking was the recognition that discretion was being provided to enable the young person to dissent from participation in the research. It can be difficult for young people to dissent in a social situation such as an outpatient clinic. The young people, if necessary, could act in a socially appropriate way to the researcher in the clinic but an option was provided to opt out if the young person did not want to participate in the study. It was the young people with arthritis who recruited the peer group that were the comparative group, and who ensured the return of the questionnaires from
the peer group. This demonstrated a high level of commitment by all of the people involved as there was a need to explain the study, gain assent and consent from the peer and the peer’s parent/guardian, complete the questionnaire, and return the questionnaires by post.

Ongoing researcher critical reflection to avoid doing harm was part of the process of the planning, conducting and evaluating of the study (Dearnley 2005).

6.5 Section Three
6.5.1 The Future – a Proposed Pathway of Care

There are a number of rheumatology models of transition including those of Sawyer and colleagues (1997) and the Vancouver model - the young adults with rheumatic disease clinic (YARD) (Tucker & Cabral 2005). Some of the models are general models of transition and primary care-based models. Rheumatology care can be complex and requires high level specific knowledge these can be barriers to general models of transitional care and current primary care based models. The most practicable model for transition is a disease focused model from paediatric services to young adult/adult rheumatology services (Tucker & Cabral 2005). Using the Ottawa Charter (1986) as a foundation stone (to build healthy public policy, create supportive environments, strengthen community action, develop personal skills and reorient health services) and acknowledging that health promotion is inextricably linked into all areas of the pathway of care, it is proposed that there needs to be an approach to care tailored to the needs of each young person with juvenile idiopathic arthritis from the onset of their disease (please see Appendix 13). The model needs to be person-centred, dynamic, and accessible in context and understanding, and it needs to capture the imagination of the young people for whom it is intended. The model proposed is one of a personal support team for the young person. It is proposed that everybody who provides care for a young person with arthritis, who educates, supports or in any other way regularly assists or befriends a young person with arthritis, take on board that they are part of that young person’s
support team. This group of people is the young person's personal strategic support team and should be formed and formalized as soon as possible following a confirmed diagnosis of JIA. Delaying the formation of this support team until the transfer from Paediatric services to Adult services will be too late. The young person with arthritis needs to understand from an early age that to successfully manage their situation they will need an excellent team around them across their lifespan, people who know and understand them and who are positively inclined towards them and their best interests. Gilligan (2000:40) refers to this as a 'scaffolding of relationships' which provides a secure base for the young person. The concept of the team is likely to be a reasonably familiar one to many young people in Ireland from as early as their primary school days. Strategic planning needs to involve all of the team including healthcare providers and the young person with arthritis. The team needs to be able to understand the current and evolving needs of the young person, it also needs to be future-focused and proactive in addressing the particular health and health promotion needs of the young person into the future.

Adapting principles from the Australian, National Chronic Disease Strategy (NHPAC 2006), the aim is to place the young person with arthritis at the centre of their support team. The team will provide support for them to optimise their own health to enable them to take increasing responsibility as they mature into adults. The model should be sufficiently flexible to capture all healthcare settings and should be applicable across the lifespan and healthcare continuum (NHPAC 2006). The proposed model of care should be sufficiently flexible to link in with Transition programmes if available. The healthcare members of the team need to be able to engage and communicate with the young person and each other on an ongoing basis. A designated care co-ordinator will be responsible for liaising with team members (please see Appendix 14). This model of care is an amalgam of models, adapted to specifically focus on young people with arthritis. Models that have been adapted to form this model are: the Chronic Care Model (Wagner et al.1996); the WHO Innovative Care for Chronic Conditions framework (WHO 2003; Epping-Jordan et al., 2004); and
The personal support team in this model may develop and evolve over time, with some members having a long and consistent involvement while others have a shorter involvement. The role of the care co-ordinator needs to be consistent for the model to function effectively. The team will appreciate that the young person needs a repertoire of coping strategies to negotiate their life pathway, as frequently JIA patients will continue to have active disease into adulthood. In some situations this sense of team may already be informally in practice. The rationale for formalising this concept of team is to enhance the young person’s sense of ownership and belonging in the team from an early age. Positive examples could include tennis stars, or the young golfer Rory McIlroy who is an extremely talented golfer but to be successful on the world stage he needs a personal support team to allow him to optimise his opportunities. Other examples include that of a racing car pit stop crew - each member with their own speciality, each member integral to the team (Please see Appendix 15) All of the crew members, in communication with the driver and each other, have the aims of optimising and maintaining to peak performance level their race car on the track. The analogy is that the young person and the young person’s support team are responsible for endeavouring to ensure that the young person is enabled to maximise their opportunities on their life track.

The aim of introducing the concept of a personal support team to young people with JIA is to encourage a sense of control of the management of their condition and to encourage pro-active self-management of their chronic condition from an early age. A person-centred model with a strong health promotion focus, the aim also, is to improve health care organization and ultimately improve health outcomes for the young people.
6.6 Summary

The quality of life of young people was protected by many factors, including friendship and social support. Social support in its many guises was a resource that assisted the young people with JIA and their families adapt to living with juvenile idiopathic arthritis. Internal and external coping strategies were developed by the young people and their families to adapt to their particular situation and were impacted by the severity of the flare-ups. The cognitive capacity to respond to disease flares was not static. Often during these times people mustered the energy to cope with their situation. Good communication skills, a sense of partnership and collaboration with the health care team were important factors in coping with the condition and improving the quality of life of the family. Parents actively encouraged socialization of their young people and maintaining peer relations even during adverse circumstances was prioritized. Parents spoke about the unknown long-term impact of the medications that were prescribed for their children. A risk-benefit analysis was done by parents; however, the burden of responsibility of their decision making caused mixed emotions and weighed heavily with the parents.

Changes in the organization of care - some of them cost-neutral, some of them small, for example asking the young people direct questions relating to their quality of life and their global well-being at each clinic visit - could make a significant impact in building relations, building communication skills, and building a global picture of the young person. Other changes in service organization, for example paediatric occupational therapy and paediatric physiotherapy, require greater resources. However, these would be a good investment in terms of enabling young people with JIA greater opportunity to optimise their life potential and to contribute to society. The American Academy of Pediatrics (2012) published a policy statement on patient and family-centered care and the Pediatrician’s role indicating its significance and benefits. Many of the recommendations and suggestions could be useful not only to Pediatricians but to other allied health care providers.
Education was seen as important to gain qualifications to attain paid income, to reduce the potential of the young person relying on long-term manual work for income, and to improve their quality of life and their long-term life prospects (Malivivya et al., 2012). Young people with the chronic disease of juvenile idiopathic arthritis need to optimise their life and health opportunities. To do this they need a flexible pathway in place to access their healthcare and a support team to assist them. The newer biologic medications have improved the quality of life of young people with arthritis, notwithstanding the unknown long-term side-effects of these medications. Reconfiguring the service organization for young people with juvenile arthritis could also be transformative. The publication of Standards of Care for juvenile idiopathic arthritis (JIA) (BSPAR 2009) a benchmark document, and related guidelines for screening for uveitis in juvenile idiopathic arthritis (BSPAR & RCOphth 2006) provide a model of best practice in the provision of care for young people with arthritis and their families. These standards have been complemented by the Arthritis and Musculoskeletal Alliance standards (ARMA) (2010) which aim to improve the quality of life of young people with JIA, using a holistic approach to care provision.

6.7 Conclusions

The quality of life of young people with the chronic disease of juvenile idiopathic arthritis in this small exploratory study was positively comparable with a peer group without JIA. Despite having difficulties, parents considered that globally their children had a good quality of life. Clinicians acknowledged the complex health issues of many young people with JIA. However, from the perspective of many years in clinical practice they were of the view that the health outcomes, functional status and quality of life of many young people with JIA had improved significantly during the course of their clinical practice life-time and were projected to improve into the future as a direct response to the availability of advancing technology, for example, genomic technology and newer medications which may provide more targeted treatment in the future (Woo & Petty 2011). These medications have been transformative in improving the quality of life of
many young people with JIA, however, the long-term outcomes of these medications are unknown, creating apprehension for families.

In general, service organization for young people with juvenile idiopathic arthritis was not integrated or person-centred. Social support was found to be a protective factor in buffering the impact of juvenile arthritis. A good team that included the young person’s family and friends, healthcare providers and educators to support them was a strong factor in adapting to and coping with the impact of JIA.

This exploratory study examined the quality of life of young people in Ireland with juvenile idiopathic arthritis from the perspectives of the young people, parents and clinicians and has contributed to the body of knowledge and scholarship related to the quality of life of young people with this condition.

In conclusion, it is important to state that since the commencement of this study there have been a number of significant and exciting developments in care provision in Ireland. At this time in 2012, a dedicated database of JIA patients has been initiated. There are two Pediatric Rheumatologists working in Ireland with a possible appointment of another in the near future. Two Chairs in Rheumatology one in University College Dublin and Trinity College Dublin have been funded from significant grants from Atlantic Philanthropies, support from the pharmaceutical industry and the Health Service Executive. The British Society for Paediatric and Adolescent Rheumatology (BSPAR) which has significant links with Irish care providers published standards of care (BSPAR 2009). These standards provide a model of care which has the potential to advance pediatric rheumatological care in Ireland exponentially.

Significant learning has taken place for the researcher relating to juvenile idiopathic arthritis and its impact on the young person and their families through discussion with clinicians and through the literature. Methodologically using a mixed methods approach required a very steep learning curve.
Interviewing people in difficult circumstances provided an opportunity to develop greater listening skills in order to optimize the potential of the interview. Interviewing the clinicians was a totally different experience to interviewing the young people and their families. These required the capacity to listen to their views but to engage them also with other views – those were particularly enjoyable to do. The importance of good communications skills so that the young people can manage and negotiate their journey with arthritis into the future provided significant learning. Putting the project together and getting it across ‘the finishing line’ has required the support of many people including academic supervisors and others who demonstrated great generosity of spirit - that too provided important learning.
6.8 Limitations of the Study

6.8.1 Methodological Issues

Mixed Methods: Critics of mixed methods research can argue that the research strategies were incompatible, with the resultant consequence of reduced value of the research.

Small Scale: This was a small exploratory study consequently the findings are unlikely to be generalizable.

Sampling: The small size of the sample of respondents of the questionnaire to young people who had JIA had the effect that the results of statistical tests that were carried out were not statistically significant.

The small number of participants available to take part in the study has implications for the reliability of the findings of the study. This is a limitation for the generalizability of the study. The sample population interviewed including young people, their parents and clinicians all came from one geographical area and may not reflect the challenges of other geographical areas.

Sequence of Interviews: The aim of the interviews with the young people was to develop questions for a health services questionnaire only. The study could have been improved theoretically if a sequence of interviews with the young people had been done at different time intervals of the project. However, it is acknowledged that access to the young people was problematic. Logistical issues including time were factors in this situation.

Flexibility: The intended course of the research needed to be adjusted as a consequence of small sample numbers. There was a need for flexibility and adaptability in order to gain other insights into the quality of life of young people with juvenile arthritis however, this could be interpreted as ‘improvisation or anything goes’ (Luckerhoff & Guillemette 2011:410).
Questionnaire: The use of a questionnaire as a data collection method has positive strengths however it can provide de-contextualized questions and variables that limit the scope of the questionnaire. The questionnaire used was developed to be age appropriate to the developmental level and cognitive level of functioning of the young people, however the researcher had reservations that the questionnaire was always sufficiently sensitive to capture the true essence of the situation.

Issues Related to Validity and Reliability: Interpretation: there is no way to be assured that the interpretation of the findings by the researcher accurately capture the true meaning of what the respondents said.

6.8.2 Other Issues

Nature of JIA: The nature of juvenile idiopathic arthritis is such that it is unpredictable and dynamic. This study provided a ‘snap shot’ of the lives of the participants on a particular day, on a different day the views of the participants could have been different.

6.9 Recommendations

The following recommendations for clinical practice and research are underpinned by the principle of recognizing the importance of focusing on the young person first and their arthritis second (Mc Donagh, 2008).

Pre-diagnosis can be a period of acute distress for families.

Recommendation: One person on the paediatric health care team to act as a co-ordinator or reference person for the young person and their family to explain the plan of action, the diagnostic procedures and results. This co-ordinator should liaise with the interagency health care team and the family.
Post-diagnosis

Communications

Post-diagnosis, it is recommended that one person on the rheumatology health care team act as a co-ordinator or reference person for the young person with arthritis and their family. The co-ordinator needs to be able to liaise with the interagency health care team and the family.

It is recommended that a model of care for young people with JIA from childhood to adulthood to meet the needs of people in different settings be developed. This could be assisted by more effective team and interagency collaboration through greater use of electronic media to provide more effective and efficient data sharing and data record keeping.

It is recommended that clinicians listen to the young people and appropriate communication strategies are utilized to allow the young people to articulate their opinions and contribute to decision making and avoid what Judd (2001:47) refers to as ‘the pleasant mask of cooperation’.

The global health of the young person their quality of life, including their health status and functional status needs to be systematically assessed at clinic appointments. A more focused approach on the global health of the young person by developing more health promoting interventions including healthy eating, exercise, and reproductive health and life skills is recommended. A more systematic approach to offering psychosocial support to a family when a child is diagnosed with juvenile arthritis could also be of significant assistance.
Medium Term Recommendations
It is recommended health provider environments including Outpatients’ Units be more child/young person friendly.
As the young people are growing up there is a need to promote a sense of transition from child to teenager. This would promote increasing responsibility, by differentiating between child clinics, youth clinics and then young adult clinics - separate times for child, youth or young adult clinics. This would assist with avoiding the reinforcement of them as children. It would acknowledge their age progression and their progressive responsibility for themselves and their condition. It is acknowledged that this is not always possible when people are travelling long distance to regional clinics but some efforts could be put in place to differentiate between the groups.

Long Term Recommendations
Strategies to promote the successful transition of young people from a paediatric system to an adult rheumatology system be implemented

Implications for Research
There is a gap in the literature relating to the quality of life of siblings of a person with JIA. Paediatric rheumatologic diseases are relatively uncommon. There is a need for large longitudinal multi-centre epidemiologic research studies to provide greater understanding of the disease and the factors that trigger the disease and the impact on quality of life.
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254


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References


References


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APPENDICES
**APPENDIX 1: Tannahill's Model of Health Promotion**

Tannahill’s model of health promotion (Downie et al. 1996) – 3 overlapping areas of activity: health education, health protection and health prevention.

Adapted from Naidoo and Wills (2000)
## Differential Diagnosis of Joint Pain in Children

<table>
<thead>
<tr>
<th>Life-threatening conditions</th>
<th>Joint pain with no joint swelling</th>
<th>Joint pain with joint swelling</th>
</tr>
</thead>
<tbody>
<tr>
<td>• Malignancy (leukaemia, lymphoma, neuroblastoma, bone tumour)</td>
<td>• Hypermobility syndromes (transient swelling is sometimes reported by patients)</td>
<td>• Juvenile idiopathic arthritis</td>
</tr>
<tr>
<td>• Sepsis (septic arthritis, osteomyelitis)</td>
<td>• Complex regional pain syndromes (localized or widespread)</td>
<td>• Trauma</td>
</tr>
<tr>
<td>• Non-accidental injury</td>
<td>• Orthopaedic syndromes (e.g. slipped upper femoral epiphysis, Perthes' disease)</td>
<td>• Infection</td>
</tr>
<tr>
<td></td>
<td>• Metabolic (e.g. hypothyroidism, lysosomal storage diseases)</td>
<td>• Septic arthritis and osteomyelitis (viral, bacterial [including Lyme disease], mycobacterial)</td>
</tr>
</tbody>
</table>

(Adapted from: Valentina, Wylie, Foster 2009)
### Appendix (5) Potential complications of juvenile idiopathic arthritis

<table>
<thead>
<tr>
<th>Chronic anterior uveitis (20%)</th>
<th>Growth disturbance</th>
<th>Constitutional problems</th>
<th>Osteoporosis</th>
<th>Macrophage activation syndrome</th>
<th>Amyloidosis</th>
<th>Psychosocial and education impact</th>
</tr>
</thead>
<tbody>
<tr>
<td>Highest risk – oligo-articular-onset JIA young female (&lt;6 year arthritis onset)</td>
<td>Generalized (short stature due to chronic disease, use of systemic corticosteroids)</td>
<td>Anaemia of chronic disease (uncommon in oligo-articular-onset JIA)</td>
<td>Multi-factorial aetiology (including reduced weight-bearing, systemic corticosteroid delay menarche and dietary factors)</td>
<td>A rare but life-threatening complication of systemic-onset JIA</td>
<td>Uncommon but high mortality – principally in severe refractory systemic-onset JIA, suggested by proteinuria</td>
<td>Depression, social isolation and unemployment are more common</td>
</tr>
<tr>
<td>Asymptomatic, potentially blinding, can be bilateral</td>
<td>Localized ('overgrown' such as leg length discrepancy due to prolonged active neck synovitis and 'undergrowth' such as micrognathia usually seen in long-standing arthritis due to premature fusion of epiphyses)</td>
<td>Fever, weight loss, malnutrition</td>
<td>Reduce risk by minimizing use of oral corticosteroids, encouraging dietary supplements of calcium and vitamin D, and regular weight-bearing exercise; consider bisphosphonates in children who sustain low trauma fractures or vertebral collapse</td>
<td>Can be triggered by infection (often viral) medication</td>
<td>Joint failure requiring joint replacement particularly patients with polyarticular disease (with or without systemic onset) and those with positive rheumatoid factor</td>
<td>Impact on the family (parents and siblings)</td>
</tr>
<tr>
<td>Detected only by slit-lamp examination and needs regular screening for several years</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

ANA – antinuclear antibodies; JIA – juvenile idiopathic arthritis  (Adapted from: Valenta, Wylie, Foster 2009)
Appendix (6) Cytokine modulators currently in use or under assessment for the treatment of severe juvenile idiopathic arthritis

<table>
<thead>
<tr>
<th>Generic name</th>
<th>Mechanism of action</th>
<th>Route of administration</th>
<th>Important issues for the child with juvenile idiopathic arthritis taking cytokine modulators</th>
</tr>
</thead>
<tbody>
<tr>
<td>Etanercept</td>
<td>TNF-α soluble receptor that binds to circulating TNF and competes with membrane receptor</td>
<td>Subcutaneous injection twice a week</td>
<td>Avoid live viral vaccines Promote annual flu vaccine Promote pneumococcal immunization (current advice 5 yearly)</td>
</tr>
<tr>
<td>Infliximab</td>
<td>Human-murine chimeric antibody that neutralizes TNF-α</td>
<td>Intravenous infusion. Various regimens, initially every 2 weeks and then every 4-6 weeks</td>
<td>Vigilance regarding infections (e.g. varicella and shingles, opportunistic infections such as listeriosis)</td>
</tr>
<tr>
<td>Adalimumab</td>
<td>Fully human monoclonal antibody that neutralizes TNF-α</td>
<td>Subcutaneous injection fortnightly</td>
<td>Advice regarding travel abroad with medicines and travel insurance</td>
</tr>
<tr>
<td>Anakinra</td>
<td>1L-1 receptor antagonist</td>
<td>Subcutaneous injection daily</td>
<td></td>
</tr>
<tr>
<td>Tocilizumab</td>
<td>1L-6 receptor antagonist</td>
<td>Intravenous infusion fortnightly or monthly</td>
<td></td>
</tr>
<tr>
<td>Abatacept</td>
<td>CTLA4-antagonist to block T-cell and B-cell interaction and initiation or the pro-inflammatory pathway</td>
<td>Given by monthly infusion</td>
<td></td>
</tr>
</tbody>
</table>

*Preparations licensed for use in juvenile idiopathic arthritis
(Adapted from: Valentina, Wylie, Foster 2009)
APPENDIX 5: Garda Clearance

An Garda Síochána

Garda Central Vetting Unit,
Technical Bureau,
Garda Headquarters,
Phoenix Park,
Dublin 8.

Date: 22 August, 2005

Strictly Private And Confidential
This document should not be construed as Proof of No Convictions, a Police Certificate, a Garda Reference or Garda Clearance.

Ms Mary O’Hara

Galway

Dear Ms O’Hara

Re: Request under Section 4 Data Protection Act 1988 & 2003

I wish to refer to your request in the above matter and to inform you that a search of the Garda Criminal Records Database was carried out.

The information which the Chief Superintendent Technical Bureau is required to give you under Section 4, Section 5 subsection 1a of the Acts is enclosed. If there are no enclosures, this means the Chief Superintendent Technical Bureau from the information you provided has been unable to reveal any personal data on you.

The searches were carried out on the basis of the identification particulars supplied to this office as per your correspondence. If you think the information enclosed, if any, is incorrect please write to the above address quoting the reference number.

I wish to acknowledge receipt of €6.35 request fee.

Yours sincerely,

[Signature]

Sergeant I/C
Central Vetting Unit
APPENDIX 6a: Ethical Approval from Hospitals

Letter removed for reasons of confidentiality and anonymity.
APPENDIX 6b: Ethical Approval from Hospitals

Letter removed for reasons of confidentiality and anonymity.
APPENDIX 6c: Ethical Approval from Hospitals

Letter removed for reasons of confidentiality and anonymity.
APPENDIX 6d: Ethical Approval from Hospitals

Letter removed for reasons of confidentiality and anonymity.
Letter removed for reasons of confidentiality and anonymity.
APPENDIX 6f: Ethical Approval from Hospitals

Letter removed for reasons of confidentiality and anonymity.
APPENDIX 6g: Ethical Approval from Hospitals

Letter removed for reasons of confidentiality and anonymity.
APPENDIX 6h: Ethical Approval from Hospitals

Letter removed for reasons of confidentiality and anonymity.
APPENDIX 7: Interview Schedule with Young People

Interviews with Young People with Jia – Interview Schedule

(a)

Topics for discussion: Interviews with young people with arthritis
Aim: to develop questions for a health services questionnaire

1. In general, have you been satisfied with your health care provision?

2. Clinic Services
   Co-ordination of services e.g. appointments
   Communications between clinicians e.g. occupational therapy and rheumatology
   Information about sources of support e.g. Arthritis Ireland,

3. Communication: Respect
   Time to listen to you?
   Your choices respected?
   The way the doctors listened to your concerns
   Understandable explanations?
   Information about treatments
   Information about medications

5. Environment
   Access to clinic staff e.g. text, telephone, email, face-to-face meeting?
   Waiting time at the clinic
   Relationship with the clinic staff
   Facilities for young people

6. Competency of clinicians
   What is good?
   How could things be made better for you?
   What is not so good?
   How could things be improved for you?

7. GP – Services
   How often do you go to the GP?
   Are you satisfied with the services? If so – what is good?
   If not – how could things be improved for you?
   Any other suggestions
### Details of Interviewees (b)

Appendix: Interviews with young people: details

**Interviews with Young People: Venue**

<table>
<thead>
<tr>
<th>Number of Interviews</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Venue: Home: Kitchen</td>
<td>06</td>
</tr>
<tr>
<td>Venue: Home: Living-room</td>
<td>02</td>
</tr>
<tr>
<td>Venue: Hotel Foyer/other</td>
<td>02</td>
</tr>
</tbody>
</table>

**Interviews with Young People: Ages of Young People**

| Aged 12-18 years | 09 |
| Aged >18 years   | 01 |

**Interviews with Young People: Parents in Attendance**

| Parent in attendance at location of interview | 09 |
| Parent not in attendance at location of interview | 01 |
APPENDIX 8: Information for Participants with Arthritis (a)

Information for Participants

Quality of Life Issues - Juvenile Idiopathic Arthritis and Young People in Ireland Study

Aim of the research study:
To find out more about the quality of life of young people in Ireland who have arthritis.

Why:
There has been very little information collected from this group of young people in Ireland.

Who:
The aim of this research is to contact all of the young people in Ireland ages 12-18 with Juvenile Idiopathic Arthritis.

How:
The information will be collected by asking young people with arthritis and young people without arthritis to complete a questionnaire. The findings will then be compared. Everything that is said will be treated in confidence and participants will not be named in any research report, presentation or paper. Participants can decide to stop being involved in the study at any time without any disadvantage to themselves.

What type of questions will be asked:
Questions will be asked about the young person’s life, for example, school, social activities, family life. The group with arthritis will be also be asked about treatment and their healthcare services.

What will be the outcome of this research:
Hopefully, more will be learnt about the issues affecting young people with this condition so that more appropriate supports can be identified and recommended.

Information about the researcher:
The researcher, Mary O’Hara, is a Nurse-Lecturer at the National University of Ireland, Galway who has a research interest in Juvenile Idiopathic Arthritis. This piece of work is towards a PhD.

What Now
You are asked to complete the white questionnaire and sign the consent form, also a Parent/Guardian must sign the consent form and return them to Mary O’Hara in the stamped addressed envelope.

You are also asked to ask a friend without arthritis to complete the yellow questionnaire and sign the consent form, also a Parent/Guardian must sign the consent form and return them to Mary O’Hara in the stamped addressed envelope provided.

Thank-you for taking the time to read this, if you have any queries please do not hesitate to ask.

Every completed & returned questionnaire is very important and will make a difference. Thanks

Contact details: Mary O’Hara, Centre for Nursing Studies, National University of Ireland, Galway.
Tel: (091) 524411 ext:3684. E-mail: mary.ohara@nuigalway.ie
Quality Of Life Questionnaire for Participants with Arthritis

(b)

Quality of life questionnaire for Adolescents

Hello,

We would like you to answer some questions about how you have been feeling during the past four weeks. These questions are about problems that young people with arthritis might have. Please answer all the questions if you can. If you don’t understand a question or would prefer not to answer it, please leave it out and go on to the next one.

- Think back over the past four weeks when answering the questions
- Choose the answer that fits you best and tick the appropriate box.

For example:

If you spend time with your friends ‘very often’ you would tick the box as shown in this example:

<table>
<thead>
<tr>
<th>Do you spend time with your friends?</th>
</tr>
</thead>
<tbody>
<tr>
<td>never</td>
</tr>
<tr>
<td>☐</td>
</tr>
</tbody>
</table>

If you make a mistake – fill in the whole square – as shown here:

<table>
<thead>
<tr>
<th>If you make a mistake - fill in the whole square - as shown here</th>
</tr>
</thead>
<tbody>
<tr>
<td>never</td>
</tr>
<tr>
<td>☐</td>
</tr>
</tbody>
</table>

There are no right or wrong answers. It’s what you think that matters.
<table>
<thead>
<tr>
<th>Part A</th>
</tr>
</thead>
</table>
| 1. Are you male or female? | ☐ female  
☐ male |
| 2. What is your date of birth? | ☐ ☐ ☐  
Day  Month  Year |
| 3. What kind of school do you attend? | ☐ National  
☐ Secondary  
☐ 6th Form College  
☐ Special School  
☐ I no longer go to school  
☐ Other ____________ |
| 4. What Class/Year year are you in at school? | ☐ 5th Class  
☐ 6th Class  
☐ 1st Year  
☐ 2nd Year  
☐ 3rd Year  
☐ Transition Year  
☐ 4th Year  
☐ 5th Year |
| 5. In which country were you born? | ☐ in Ireland  
☐ in a different country  
Which one? |
| 6. In which country was your mother born? | ☐ in Ireland  
☐ in a different country  
Which one? |
<table>
<thead>
<tr>
<th>Question</th>
<th>Options</th>
</tr>
</thead>
<tbody>
<tr>
<td>7. In which country was your father born?</td>
<td>□ in Ireland</td>
</tr>
<tr>
<td></td>
<td>□ in a different country</td>
</tr>
<tr>
<td></td>
<td>Which one?</td>
</tr>
<tr>
<td>8. How well off do you think your family is?</td>
<td>□ very well off</td>
</tr>
<tr>
<td></td>
<td>□ quite well off</td>
</tr>
<tr>
<td></td>
<td>□ average</td>
</tr>
<tr>
<td></td>
<td>□ not very well off</td>
</tr>
<tr>
<td></td>
<td>□ not at all well off</td>
</tr>
<tr>
<td>9. Does your family own a car, van or truck?</td>
<td>□ no</td>
</tr>
<tr>
<td></td>
<td>□ yes</td>
</tr>
<tr>
<td></td>
<td>□ yes, two or more</td>
</tr>
<tr>
<td>10. Do you have your own bedroom?</td>
<td>□ no</td>
</tr>
<tr>
<td></td>
<td>□ yes</td>
</tr>
<tr>
<td>11. During the past 12 months, how many times did you travel away on holiday with your family?</td>
<td>□ not at all</td>
</tr>
<tr>
<td></td>
<td>□ once</td>
</tr>
<tr>
<td></td>
<td>□ twice</td>
</tr>
<tr>
<td></td>
<td>□ more than twice</td>
</tr>
<tr>
<td>12. How many computers does your family own?</td>
<td>□ none</td>
</tr>
<tr>
<td></td>
<td>□ one</td>
</tr>
<tr>
<td></td>
<td>□ two</td>
</tr>
<tr>
<td></td>
<td>□ more than two</td>
</tr>
<tr>
<td>13. How much do you weigh (without clothes)? *</td>
<td>□ □ □ pounds</td>
</tr>
<tr>
<td>14. How tall are you (without shoes)? *</td>
<td>□ feet □ inches</td>
</tr>
</tbody>
</table>

(* A guess will do if you’re not sure)
### Part B

<table>
<thead>
<tr>
<th>Question</th>
<th>never</th>
<th>seldom</th>
<th>quite often</th>
<th>very often</th>
<th>always</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Are you confident about your future?</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
<tr>
<td>2. Do you enjoy your life?</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
<tr>
<td>3. Are you able to do everything you want to do even though you have arthritis?</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
<tr>
<td>4. Do you feel like everyone else even though you have arthritis?</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
<tr>
<td>5. Are you free to lead the life you want even though you have arthritis?</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
<tr>
<td>6. Are you able to do things without your parents?</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
<tr>
<td>7. Are you able to run and move as you like?</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
<tr>
<td>8. Do you feel tired because of your arthritis?</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
<tr>
<td>9. Is your life ruled by your arthritis?</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
<tr>
<td>10. Does it bother you that you have to explain to others what you can and can’t do?</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
<tr>
<td></td>
<td>Question</td>
<td>never</td>
<td>seldom</td>
<td>quite often</td>
<td>very often</td>
</tr>
<tr>
<td>---</td>
<td>-------------------------------------------------------------------------</td>
<td>-------</td>
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<td>------------</td>
</tr>
<tr>
<td>11</td>
<td>Is it difficult to sleep because of your arthritis?</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>12</td>
<td>Does your arthritis bother you when you play?</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>13</td>
<td>Does your arthritis make you feel bad about yourself?</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>14</td>
<td>Are you unhappy because you have arthritis?</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>15</td>
<td>Do you worry about your arthritis?</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>16</td>
<td>Does your arthritis make you angry?</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>17</td>
<td>Do you have fears about the future because of your arthritis?</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>18</td>
<td>Does your arthritis get you down?</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>19</td>
<td>Does it bother you that your life has to be planned?</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>20</td>
<td>Do you feel lonely because of your arthritis?</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Question</td>
<td>never</td>
<td>seldom</td>
<td>quite often</td>
<td>very often</td>
</tr>
<tr>
<td>---</td>
<td>--------------------------------------------------------------------------</td>
<td>-------</td>
<td>--------</td>
<td>-------------</td>
<td>------------</td>
</tr>
<tr>
<td>21</td>
<td>Do your teachers behave differently towards you than towards others?</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>22</td>
<td>Do you have problems concentrating at school because of your arthritis?</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>23</td>
<td>Do you feel that others have something against you?</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>24</td>
<td>Do you think that others stare at you?</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>25</td>
<td>Do you feel different from other children?</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>26</td>
<td>Do other kids understand about your arthritis?</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>27</td>
<td>Do you go out with your friends?</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>28</td>
<td>Are you able to play or do things with other children/adolescents (e.g. sports)?</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>29</td>
<td>Do you think that you can do most things as well as other children?</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>30</td>
<td>Do your friends enjoy being with you?</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>31</td>
<td>Do you find it easy to talk about your arthritis to other people?</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
# About your medical treatment

Do you take any medicine for your arthritis?

- [ ] yes  
- [ ] no

If yes, please fill in the following questions

If no, then skip this section and go to section C

<table>
<thead>
<tr>
<th>Think about the past four weeks...</th>
<th>never</th>
<th>seldom</th>
<th>quite often</th>
<th>very often</th>
<th>always</th>
</tr>
</thead>
<tbody>
<tr>
<td>32 Does having to get help with medication from others bother you?</td>
<td>❑</td>
<td>❑</td>
<td>❑</td>
<td>❑</td>
<td>❑</td>
</tr>
<tr>
<td>33 Is it annoying for you to have to remember your medication?</td>
<td>❑</td>
<td>❑</td>
<td>❑</td>
<td>❑</td>
<td>❑</td>
</tr>
<tr>
<td>34 Are you worried about your medication?</td>
<td>❑</td>
<td>❑</td>
<td>❑</td>
<td>❑</td>
<td>❑</td>
</tr>
<tr>
<td>35 Does taking medication bother you?</td>
<td>❑</td>
<td>❑</td>
<td>❑</td>
<td>❑</td>
<td>❑</td>
</tr>
<tr>
<td>36 Do you hate taking your medicine?</td>
<td>❑</td>
<td>❑</td>
<td>❑</td>
<td>❑</td>
<td>❑</td>
</tr>
<tr>
<td>37 Does taking medication disrupt everyday life?</td>
<td>❑</td>
<td>❑</td>
<td>❑</td>
<td>❑</td>
<td>❑</td>
</tr>
</tbody>
</table>
Part C

Now we would like to ask you about your arthritis. The first three questions are about how much trouble you have had with your arthritis in the last year.

Please tick the appropriate box (√)

<table>
<thead>
<tr>
<th>In the last year</th>
<th>never</th>
<th>a few</th>
<th>every</th>
<th>every</th>
<th>daily</th>
</tr>
</thead>
<tbody>
<tr>
<td>A How often did you have problems with your arthritis?</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Not at all</th>
<th>a little bit</th>
<th>moderately</th>
<th>quite a bit</th>
<th>extremely</th>
</tr>
</thead>
<tbody>
<tr>
<td>B How severe was your arthritis during the last year?</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>never</th>
<th>seldom</th>
<th>quite often</th>
<th>very often</th>
<th>always</th>
</tr>
</thead>
<tbody>
<tr>
<td>C How often did you have pain in your joints or muscles</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
<tr>
<td></td>
<td>Question</td>
<td>never</td>
<td>seldom</td>
<td>quite often</td>
</tr>
<tr>
<td>---</td>
<td>--------------------------------------------------------------------------</td>
<td>-------</td>
<td>--------</td>
<td>-------------</td>
</tr>
<tr>
<td>1</td>
<td>Do you get exhausted easily?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>2</td>
<td>Does arthritis make you feel too exhausted to be with friends?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>3</td>
<td>Do you hate being in pain?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>4</td>
<td>Does it annoy you that the pain sometimes comes on so suddenly?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>5</td>
<td>Does pain stop you from doing what you want?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>6</td>
<td>Does it bother you that you can’t do all sports/hobbies because of your arthritis?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>7</td>
<td>Do you hate being restricted in movement?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>8</td>
<td>Does it bother you that you have trouble writing/drawing?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>9</td>
<td>Do others understand that your symptoms may change suddenly?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>10</td>
<td>Do your friends understand that you may feel poorly quite suddenly?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>11</td>
<td>Do teachers understand that you sometimes can’t join in?</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
### Part D

<table>
<thead>
<tr>
<th></th>
<th>Question</th>
<th>Scale</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>In general, how would you say your health is?</td>
<td>excellent</td>
</tr>
<tr>
<td>2</td>
<td>Have you felt fit and well?</td>
<td>not at all</td>
</tr>
<tr>
<td>3</td>
<td>Have you been physically active (e.g., running, climbing and biking)?</td>
<td>not at all</td>
</tr>
<tr>
<td>4</td>
<td>Have you been able to run well?</td>
<td>never</td>
</tr>
<tr>
<td>5</td>
<td>Have you felt full of energy?</td>
<td>not at all</td>
</tr>
<tr>
<td>6</td>
<td>Has your life been enjoyable?</td>
<td>never</td>
</tr>
<tr>
<td>7</td>
<td>Have you been in a good mood?</td>
<td>never</td>
</tr>
<tr>
<td>8</td>
<td>Have you had fun?</td>
<td>never</td>
</tr>
<tr>
<td>9</td>
<td>Have you felt sad?</td>
<td>never</td>
</tr>
<tr>
<td>10</td>
<td>Have you felt so bad that you didn't want to do anything?</td>
<td>never</td>
</tr>
<tr>
<td></td>
<td>Question</td>
<td>never</td>
</tr>
<tr>
<td>---</td>
<td>--------------------------------------------------------------------------</td>
<td>-------</td>
</tr>
<tr>
<td>11</td>
<td>Have you felt lonely?</td>
<td>❑</td>
</tr>
<tr>
<td>12</td>
<td>Have you been happy with the way you are?</td>
<td>❑</td>
</tr>
<tr>
<td>13</td>
<td>Have you had enough time for yourself?</td>
<td>❑</td>
</tr>
<tr>
<td>14</td>
<td>Have you been able to do the things you want to do in your free time?</td>
<td>❑</td>
</tr>
<tr>
<td>15</td>
<td>Have your parent(s) had enough time for you?</td>
<td>❑</td>
</tr>
<tr>
<td>16</td>
<td>Have your parents treated you fairly?</td>
<td>❑</td>
</tr>
<tr>
<td>17</td>
<td>Have you been able to talk to your parent(s) when you wanted to?</td>
<td>❑</td>
</tr>
<tr>
<td>18</td>
<td>Have you had enough money to do the same things as your friends?</td>
<td>❑</td>
</tr>
<tr>
<td>19</td>
<td>Have you had enough money for your expenses?</td>
<td>❑</td>
</tr>
<tr>
<td>20</td>
<td>Have you spent time with your friends?</td>
<td>❑</td>
</tr>
<tr>
<td>21</td>
<td>Have you had fun with your friends?</td>
<td>❑</td>
</tr>
<tr>
<td>22</td>
<td>Have you and your friends helped each other?</td>
<td>❑</td>
</tr>
<tr>
<td>23</td>
<td>Have you been able to rely on your friends?</td>
<td>❑</td>
</tr>
<tr>
<td></td>
<td>Think about the past four weeks...</td>
<td>not at all</td>
</tr>
<tr>
<td>---</td>
<td>----------------------------------</td>
<td>------------</td>
</tr>
<tr>
<td>24</td>
<td>Have you been happy at school?</td>
<td></td>
</tr>
<tr>
<td>25</td>
<td>Have you got on well at school?</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>never</td>
</tr>
<tr>
<td>26</td>
<td>Have you been able to pay</td>
<td></td>
</tr>
<tr>
<td></td>
<td>attention?</td>
<td></td>
</tr>
<tr>
<td>27</td>
<td>Have you got along well with your</td>
<td></td>
</tr>
<tr>
<td></td>
<td>teachers?</td>
<td></td>
</tr>
</tbody>
</table>
### Part E

Please tick one box on each line (√)

<table>
<thead>
<tr>
<th>Please think about your health care services</th>
<th>excellent</th>
<th>very good</th>
<th>good</th>
<th>fair</th>
<th>poor</th>
</tr>
</thead>
<tbody>
<tr>
<td>1 In general, how would you say your health care services are?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th></th>
<th>never</th>
<th>seldom</th>
<th>quite often</th>
<th>very often</th>
<th>always</th>
</tr>
</thead>
<tbody>
<tr>
<td>2 Does your outpatient clinic doctor listen to your views?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>3 Does your GP listen to your views?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>4 Do you have to wait more than two hours at routine outpatient clinics?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>5 Has there been bruising or swelling in your hand or arm where you have had blood taken?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>6 Do you have to travel more than forty miles /64 km (approx) to your arthritis outpatient clinic appointment?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Q 7-10 Please tick one box on each line</td>
<td>not at all</td>
<td>slightly</td>
<td>moderately</td>
<td>very</td>
<td>extremely</td>
</tr>
<tr>
<td>--------------------------------------</td>
<td>------------</td>
<td>----------</td>
<td>------------</td>
<td>------</td>
<td>-----------</td>
</tr>
<tr>
<td>7 Would more information about your arthritis be helpful for you?</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
<tr>
<td>8 Would more information for your teachers about juvenile arthritis be helpful?</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
<tr>
<td>9 Would more information for your friends about juvenile arthritis be helpful?</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
<tr>
<td>10 Would it be helpful if you also met other health professionals at your routine arthritis outpatient appointments?</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
<tr>
<td>11 Please tick (+) which health care people it would be most helpful to meet. (Please tick more than one box if appropriate)</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
<tr>
<td></td>
<td>physiotherapist</td>
<td>occupational therapist</td>
<td>clinical nurse specialist</td>
<td>eye doctor</td>
<td>other, please specify</td>
</tr>
<tr>
<td>12 Would it be helpful if your medications were free?</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
<tr>
<td>13 Would 'on line' prescriptions from your doctor to your pharmacist be helpful?</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
<tr>
<td>14 Would meeting young people your own age with arthritis be helpful</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
<tr>
<td>15 Do you feel that you are treated with respect by healthcare professionals?</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
<tr>
<td>Please tick one box on each line</td>
<td>high level of benefit</td>
<td>moderate benefit</td>
<td>slight benefit</td>
<td></td>
<td></td>
</tr>
<tr>
<td>----------------------------------</td>
<td>-----------------------</td>
<td>------------------</td>
<td>----------------</td>
<td></td>
<td></td>
</tr>
<tr>
<td>16 Which would be of benefit to you?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>More information for the general public about juvenile arthritis</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Free medication</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Only very experienced people to take blood</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Greater respect from Healthcare professionals</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Doctors listening to my views</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td></td>
<td></td>
</tr>
<tr>
<td>On-line prescriptions</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td></td>
<td></td>
</tr>
<tr>
<td>One-stop shop clinics</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td></td>
<td></td>
</tr>
<tr>
<td>One set of patient records that all Healthcare professionals work from.</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Single contact person who is responsible for co-ordination between departments for appointments of treatments</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Other suggestions: please state</td>
<td>☐</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Thank you very much for helping us with the study. Your answers will help us to find out more about the lives of young people with arthritis!
Information for Participants without Arthritis (c)

Information to Participants without Juvenile Idiopathic Arthritis  Version: 2 Date: 06/07/02

Quality of Life Issues – Juvenile Idiopathic Arthritis and Young People in Ireland Study

Aim of the research study:
To find out more about the quality of life of young people in Ireland who have arthritis.

Why:
There has been very little information collected from this group of young people in Ireland.

Who:
The aim of this research is to contact all of the young people in Ireland ages 12-18 with Juvenile Idiopathic Arthritis and compare the findings with a group of young people who do not have arthritis. You are being asked to be part of the group that does not have arthritis.

How:
Young people without arthritis will be asked to complete a questionnaire (yellow) and sign a consent form. Also their parent/guardian must sign a consent form. Everything that is said will be treated in confidence and participants will not be named in any research report, presentation or paper. Participants can decide to stop being involved in the study at any time without any disadvantage to themselves.

What type of questions will be asked:
Questions will be asked about the young person’s life, for example, school, social activities, family life. The group with arthritis will also be asked about treatment and their health care services.

What will be the outcome of this research:
Hopefully, more will be learnt about the issues affecting young people with arthritis so that more appropriate supports can be identified and recommended.

Information about the researcher:
The researcher, Mary O’Hara, is a Nurse-Lecturer at the National University of Ireland, Galway who has a research interest in Juvenile Idiopathic Arthritis. This piece of work is towards a PhD.

What Now:
You are asked to complete the yellow questionnaire and sign the consent form, also a parent/guardian must sign the consent form and return them to Mary O’Hara in the stamped addressed envelope.

Thank-you for taking the time to read this, if you have any queries please do not hesitate to ask.

Every completed & returned questionnaire is very important and will make a difference. Thank-you

Contact details: Mary O’Hara, Centre for Nursing Studies, National University of Ireland, Galway. Tel: (091) 524411 ext:3684 e-mail: mary.ohara@nuigalway.ie
Quality Of Life Questionnaire for Participants without Arthritis

Quality of life questionnaire for Adolescents

Hello,

We would like you to answer some questions about how you have been feeling during the past four weeks. These questions relate to young people that do not have arthritis and young people with arthritis. Please answer all the questions if you can. If you don't understand a question or would prefer not to answer it, please leave it out and go on to the next one.

- Think back over the past four weeks when answering the questions
- Choose the answer that fits you best and tick the appropriate box.

For example:

If you spend time with your friends 'very often' you would tick the box as shown in this example:

<table>
<thead>
<tr>
<th>Do you spend time with your friends?</th>
<th>never</th>
<th>seldom</th>
<th>quite often</th>
<th>very often</th>
<th>always</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>✔</td>
<td>☐</td>
</tr>
</tbody>
</table>

If you make a mistake - fill in the whole square - as shown here

<table>
<thead>
<tr>
<th>If you make a mistake - fill in the whole square - as shown here</th>
<th>never</th>
<th>seldom</th>
<th>quite often</th>
<th>very often</th>
<th>always</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
<td>☐</td>
</tr>
</tbody>
</table>

There are no right or wrong answers. It's what you think that matters.
| 1. Are you male or female? | ☐ female  
|                            | ☐ male |
| 2. What is your date of birth? | ☐ ☐ ☐ ☐   
|                              | Day Month Year |
| 3. What kind of school do you attend? | ☐ National  
|                                          | ☐ Secondary  
|                                          | ☐ 6th Form College  
|                                          | ☐ Special School  
|                                          | ☐ I no longer go to school  
|                                          | ☐ Other ____________ |
| 4. What Class/Year year are you in at school? | ☐ 5th Class  
|                                          | ☐ 6th Class  
|                                          | ☐ 1st Year  
|                                          | ☐ 2nd Year  
|                                          | ☐ 3rd Year  
|                                          | ☐ Transition Year  
|                                          | ☐ 4th Year  
|                                          | ☐ 5th Year  
| 5. In which country were you born? | ☐ in Ireland  
|                                          | ☐ in a different country  
|                                          | Which one? |
| 6. In which country was your mother born? | ☐ in Ireland  
|                                          | ☐ in a different country  
<p>|                                          | Which one? |</p>
<table>
<thead>
<tr>
<th>Question</th>
<th>Options</th>
</tr>
</thead>
<tbody>
<tr>
<td>7. In which country was your father born?</td>
<td>in Ireland in a different country Which one?</td>
</tr>
<tr>
<td>8. How well off do you think your family is?</td>
<td>very well off quite well off average not very well off not at all well off</td>
</tr>
<tr>
<td>9. Does your family own a car, van or truck?</td>
<td>no yes yes, two or more</td>
</tr>
<tr>
<td>10. Do you have your own bedroom?</td>
<td>no yes</td>
</tr>
<tr>
<td>11. During the past 12 months, how many times did you travel away on holiday with your family?</td>
<td>not at all once twice more than twice</td>
</tr>
<tr>
<td>12. How many computers does your family own?</td>
<td>none one two more than two</td>
</tr>
<tr>
<td>13. How much do you weigh (without clothes)? *</td>
<td>pounds</td>
</tr>
<tr>
<td>14. How tall are you (without shoes)? *</td>
<td>feet inches</td>
</tr>
</tbody>
</table>

(* A guess will do if you're not sure)
Think about the past four weeks...
Please tick one box on each line

<p>| | | | | |</p>
<table>
<thead>
<tr>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>In general, how would you say your health is?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>2</td>
<td>Have you felt fit and well?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>3</td>
<td>Have you been physically active (e.g. running, climbing and biking)?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>4</td>
<td>Have you been able to run well?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>5</td>
<td>Have you felt full of energy?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>6</td>
<td>Has your life been enjoyable?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>7</td>
<td>Have you been in a good mood?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>8</td>
<td>Have you had fun?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>9</td>
<td>Have you felt sad?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>10</td>
<td>Have you felt so bad that you didn't want to do anything?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>never</td>
<td>seldom</td>
<td>quite often</td>
</tr>
<tr>
<td>---</td>
<td>-----------------------------------------------------------------</td>
<td>-------</td>
<td>--------</td>
<td>-------------</td>
</tr>
<tr>
<td>11</td>
<td>Have you felt lonely?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>12</td>
<td>Have you been happy with the way you are?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>13</td>
<td>Have you had enough time for yourself?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>14</td>
<td>Have you been able to do the things you want to do in your free time?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>15</td>
<td>Have your parent(s) had enough time for you?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>16</td>
<td>Have your parents treated you fairly?</td>
<td></td>
<td></td>
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</tr>
<tr>
<td>17</td>
<td>Have you been able to talk to your parent(s) when you wanted to?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>18</td>
<td>Have you had enough money to do the same things as your friends?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>19</td>
<td>Have you had enough money for your expenses?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>20</td>
<td>Have you spent time with your friends?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>21</td>
<td>Have you had fun with your friends?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>22</td>
<td>Have you and your friends helped each other?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>23</td>
<td>Have you been able to rely on your friends?</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
Think about the past four weeks...
Please tick one box on each line

<table>
<thead>
<tr>
<th></th>
<th></th>
<th>not at all</th>
<th>slightly</th>
<th>moderately</th>
<th>very</th>
<th>extremely</th>
</tr>
</thead>
<tbody>
<tr>
<td>24</td>
<td>Have you been happy at school?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>25</td>
<td>Have you got on well at school?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>26</td>
<td>Have you been able to pay attention?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>27</td>
<td>Have you got along well with your teachers?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Thank you very much for helping with the study.
## APPENDIX 9: Interview Schedule for Parents

### Semi-Structured Interviews with Parents - Schedule

<table>
<thead>
<tr>
<th>Number</th>
<th>Theme</th>
<th>Concept</th>
<th>Question</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Introductions</td>
<td></td>
<td></td>
</tr>
<tr>
<td>2</td>
<td>Introduce condition</td>
<td>Present problem</td>
<td></td>
</tr>
<tr>
<td>3</td>
<td>Understanding nature of condition</td>
<td></td>
<td></td>
</tr>
<tr>
<td>4</td>
<td>Coping</td>
<td>Coping</td>
<td>What was it like for you when your child was newly diagnosed with JIA?</td>
</tr>
<tr>
<td>5</td>
<td>Family</td>
<td>Stressors</td>
<td>Could you tell us what a good day is like for you?</td>
</tr>
<tr>
<td>6</td>
<td>Family environment</td>
<td>Impact on the family</td>
<td>Could you tell us what a bad day is like for you?</td>
</tr>
<tr>
<td>7</td>
<td>Child’s adjustment</td>
<td>Support</td>
<td>What did you talk about with your child’s condition and are in a state of ‘high risk’?</td>
</tr>
<tr>
<td>8</td>
<td>Knowledge/Perception</td>
<td>Compromises</td>
<td>Other parents talk how they are constantly monitoring child’s condition and are in a state of ‘high risk’</td>
</tr>
<tr>
<td>9</td>
<td>Adolescents</td>
<td>Adolescent issues</td>
<td>How have you found the best way to cope with your child’s condition</td>
</tr>
<tr>
<td>10</td>
<td>Complexity of care</td>
<td>What have you found to be the most difficult thing for you in caring for your child with JIA?</td>
<td></td>
</tr>
<tr>
<td>11</td>
<td>Support network</td>
<td>Support network</td>
<td>What supports (e.g., family/friends) have you found most helpful?</td>
</tr>
<tr>
<td>12</td>
<td>Management of condition</td>
<td>How do you keep track of appointments with health care providers?</td>
<td></td>
</tr>
<tr>
<td>13</td>
<td>Quality of care</td>
<td>management</td>
<td>How would you rate the quality of care that your child is receiving?</td>
</tr>
<tr>
<td>14</td>
<td>Transition to adult services</td>
<td>Transition to adult services</td>
<td>How has your child been able to transition from your child’s condition?</td>
</tr>
<tr>
<td>15</td>
<td>Other</td>
<td>Other</td>
<td>Where and when was the transition from pediatric to adult care?</td>
</tr>
<tr>
<td>16</td>
<td>Recommendations for the future</td>
<td>Recommendations for the future</td>
<td>What do you recommend for others in similar situations?</td>
</tr>
<tr>
<td>17</td>
<td>Any other issues</td>
<td>Any other issues</td>
<td>Preparation for a young person</td>
</tr>
</tbody>
</table>

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**Page 1-1**  
**Page 2-2**  
**Page 3-3**  
**Page 4-4**
APPENDIX 10:  Topics for Discussion with Clinicians

Topics for discussions with Clinicians

1. The role of the multidisciplinary team/ cost effectiveness
   Communication and decision-making
   Cause and course of disease: Are you asked about the cause, course
   and treatment of JIA

2. Control of disease
   Co-morbidities – side-effects of medications
   Uveitis
   Do they comply with treatment management
   If not, why not?

3. Adherence/compliance
   Pain management
   Disease education
   Exercise
   Recreation and play
   Physiotherapy and hydrotherapy /aerobic fitness
   Orthotics
   Physical growth
   Nutrition
   Bone density
   Dental health
   General health issues
   Monitoring of progress
   Emerging therapies

4. Transitional care
   Education and vocation – the big picture

5. The future: Access to care
   What are you proposing into the future re: alternative and innovative
   ways of providing care?
   How acute care services differ from chronic care services?
   Community-based teams?
   What constitutes optimal services?
APPENDIX 11: Consultant Survey

(a)

Juvenile Idiopathic Arthritis and Young People in Ireland – A Quality of Life Study

Survey to Paediatricians and Rheumatologists in Ireland

1. Do you have any patients with Juvenile Idiopathic Arthritis (JIA)?
   (please tick the appropriate box)

   No □
   1-10 patients □
   11-20 patients □
   More than 20 patients □

2. How many patients do you have with Juvenile Idiopathic Arthritis aged 12-18?
   (please tick the appropriate box)

   None □
   1-10 patients □
   11-20 patients □
   More than 20 patients □
Letter to Consultants (b)

30 November 2004

Dear

Re:– Summary of results of Survey to Consultant Paediatricians and Rheumatologists part of a PhD Study: Quality of Life Issues: Juvenile Idiopathic Arthritis and Young People in Ireland

Please find enclosed a summary of results of the above study.

I would like to take this opportunity to thank you for your participation, as well as helpful and supportive comments which were very encouraging to a student. The response rate of the study was 77%. Also, 50% were completed and returned within one week of being sent out. In this simple survey it is important to acknowledge that there could be an overlap in the statistics of some patients attending both, a Paediatrician and a Rheumatologist. It is expected that this issue will be resolved before the study is completed. The names and addresses of Paediatricians and Rheumatologists were obtained from the Irish Medical Directory 2003 and 2004.

The last phase of my data collection is to use a condition specific quality of life questionnaire and a service provision questionnaire to ascertain if/how arthritis impacts on the young person’s life. These questionnaires can be completed by the young person or their parent/guardian. It is anticipated that I will be in contact with you again early in the new year if you have young people aged between 12-18 with Juvenile Arthritis.

If you have any queries or any comments they would all be welcomed.

Again, very many thanks.

Nollaig shonasach agus athbhliain shuaimhneach

____________________________
Mary O’Hara
Tel: 091 493684
e.mail: mary.ohara@nuigalway.ie
Summary of Results of Survey to Consultant Paediatricians and Rheumatologists (c)

Summary of Results of Survey to Consultant Paediatricians and Rheumatologists in Ireland - November 2004

This survey is part of a PhD Study, 'Quality of Life Issues: Juvenile Idiopathic Arthritis and Young People in Ireland', National University of Ireland, Galway

Survey Aim: To ascertain how many Consultant Paediatricians and Rheumatologists are caring for young people with Juvenile Idiopathic Arthritis in the Republic of Ireland

Survey Response

Survey Question 1:
Do you have any patients with Juvenile Idiopathic Arthritis (JIA)

Survey Question 2:
How many patients do you have with JIA aged 12-18?

Mary O’Flataí, PhD Study, Quality of Life Issues: Juvenile Idiopathic Arthritis and Young People in Ireland, National University of Ireland, Galway, Tel: 091 495884 e-mail: mary.oflatai@nuigalway.ie
APPENDIX 12: Data from Three Perspectives

The purpose of the diagram is to demonstrate the collection of data from multiple perspectives relating to the health-related quality of life of young people with juvenile idiopathic arthritis. The diagram represents the uniqueness of the data from each group but also indicates areas of commonality within the groups demonstrated by the circles overlapping.
APPENDIX 13: An Example of Physical and Personal Issues to Emerge From Interviews with Parents (a)
Key Issues to Emerge: Young People, Parents and Clinicians

(b)
Resources to Manage JIA – Stepping Stone Approach (c)

An Example of physical and personal resources to manage JIA – Stepping stone approach

- **Personal resources**
  - e.g. psychosocial skills
  - Resilience
  - Coping strategies

- **Disease management regimen**
  - Rheum Specialist team
  - GP/Nurse
  - Physio
  - Ophthalmologist
  - OT

- **Communication**
  - Collaboration/partnership
  - Inter-sectoral communication
  - Informatics systems

- **Social Support**
- **IT**
- **Friends**
APPENDIX 14: An Example of Coping Resources/Skills Needed By a Young Person with JIA
APPENDIX 15: Analogy of the Pit Stop Crew

The Team Work Collaboratively To Optimize the Performance of the Driver and the Car

Image removed for copyright reasons.
**Glossary**

**Active arthritis**\(^1\)
Swelling within a joint, or limitation in range of joint movement with joint pain or tenderness.

**Clinical remission**\(^1\)

*Clinical remission on medication:* The criteria for inactive disease must be met for a minimum of 6 continuous months while the patient is on medication in order for the patient to be considered to be in a state of clinical remission on medication.

*Clinical remission off medication:* The criteria for inactive disease must be met for a minimum of 12 continuous months while off all anti-arthritis and anti-uveitis medications in order for the patient to be considered to be in a state of clinical remission off medication.

‘Discursive othering’
The use of parents’ views on their children’s wants and needs as a proxy for the child’s own voice (Dowse 2001).

**Flare**\(^1\)
Any increase in disease activity in a child with previous or current arthritis which requires and increase in treatment. Although a flare of disease does not necessarily lead to a change in treatment, any clinically significant flare should.
Disease Activity

*Mild disease*
Active arthritis which:

1. Does not interfere with a child's normal activities of daily living.
2. Is not associated with any evidence of disease progression (either clinically or radiographically) over a 6-month period.

*Moderate disease*
Active arthritis which:

1. Interferes with a child’s normal activities to some extent, but does not prevent participation in most school activities including physical education.
2. Apparently mild disease, but associated with disease progression (either clinically or radiographically).

*Severe disease*

1. Active arthritis which severely inhibits normal activities of daily living.
2. Active arthritis associated with the development of erosions and/or ankylosis
An example of a ‘micro-context’ model of care for young people with JIA. Implicit in the model is the concept of health promotion. The care framework needs to be user-friendly and to meet the needs of the person with JIA across their lifespan and across different health settings. Juvenile idiopathic arthritis is an unpredictable disease. The care framework needs to facilitate fluctuations, early disease, established disease and refractory disease. The disease trajectory is dynamic – need to be able to ‘touch back’ to certain members of the team for modification, e.g. treatment, education. Pathway – unique to the individual – easy access is important. Assists person with JIA (and parent/guardian) to build up a personal care team providing, knowledge, treatment and support through paediatric service provision to adult care provision.