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Healthcare 'Fit' and autism: An examination of barriers to, and experiences of, physical healthcare for people on the autism spectrum

A thesis submitted to the School of Medicine, National University of Ireland Galway in fulfilment of the requirements for the Degree of Doctor of Philosophy (Population Health and Health Services Research)
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Declaration

This work is submitted to fulfil the requirement of the degree of Doctor of Philosophy, at the National University of Ireland, Galway. No part of this thesis has been previously submitted at this or at any other university. There is a declaration at the beginning of each chapter to specify my contribution to each study. I declare that this thesis is entirely my own work.

Signed: Date: 29/06/2021

Chloe Walsh

Summary of Thesis

Autistic individuals experience substantial health inequities, reflected in poorer health outcomes and higher mortality rates. One suggested determinant of this health inequity is issues in access to healthcare. This thesis, therefore, aimed to examine the barriers to healthcare for autistic individuals and consider how access might be improved.

Five empirical studies were completed. Study 1 comprised a systematic review of barriers to healthcare reported by autistic individuals, caregivers, and healthcare providers (HCPs). A taxonomy of barriers was developed comprising four themes: barriers associated with autism-related characteristics; other patient-related barriers; HCP-related barriers; and system-related barriers.

Study 2 described the development and preliminary evaluation of a novel caregiver-report tool to assess barriers to care, which consisted of four factors: patient-related barriers, HCP-related barriers, system-related barriers, and barriers related to managing care. The most frequently occurring barriers included difficulties identifying or reporting pain/symptoms and a lack of HCP knowledge about autism.

Study 3 described the development and preliminary evaluation of a physician-report tool to assess barriers to providing care to autistic individuals, which consists of three factors: patient-related barriers; HCP/family-related barriers, and system-related barriers. The most common barriers included insufficient patient supports, and communication difficulties.

Study 4 describes the use of patient narratives to identify barriers occurring in challenging healthcare encounters for autistic individuals and assessed the impact these had on patients. Patient-related barriers occurred most often, followed by HCP-related barriers. More than a quarter of the described encounters were rated as high severity.

Study 5 presents a systematic review of interventions aimed at improving access to, or experiences in, healthcare for autistic individuals. Interventions were mostly patient-focused with fewer studies targeting the HCP or the system.

The data presented herein demonstrate that autistic individuals face substantial health inequities. Thus, models of healthcare must change to ensure optimal health for the entire autistic community.

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Finally, I want to express a very special thank you to all my participants. I am so grateful to you for taking the time to complete surveys and to share your stories.

Dedication

I dedicate this thesis to my parents, Marie and Pete, for instilling in me from a young age, the importance advocating for the rights persons with disabilities and for leading by example through their work. I also dedicate this to my grandmother, Eithne (the first Dr Walsh), who taught me the importance of education and independence, especially for women.

Finally, I dedicate this thesis to my participants who were so generous with their time and experiences. Without you, this thesis would not have been possible.

Publications and Conference Proceedings Arising from this Thesis

Publications:

1. Walsh C, Lydon S, O'Dowd E, O'Connor P. Barriers to healthcare for persons with autism: A systematic review of the literature and development of a taxonomy. Dev Neurorehabil. 2020 Oct 2;23(7):413-30. doi:10.1080/17518423.2020.1716868

2. Walsh C, Lydon S, Hehir A, O'Connor P. Development and evaluation of a novel caregiver-report tool to assess barriers to physical healthcare for people on the autism spectrum. Res Autism Spect Disord. 2020 Nov 1;79:101680. doi:10.1016/j.rasd.2020.101680

3. Walsh C, Lydon S, Walsh E, O'Connor P. A systematic review of interventions to improve healthcare experiences and access in Autism. Rev J Autism Dev Disabil. 2021 Aug 26; https://doi.org/10.1007/s40489-021-00279-2

4. Walsh C, Lydon S, Geoghegan R, Carey C, Creed M, O'Loughlin L, Walsh E, Byrne D, O'Connor, P. Development and preliminary evaluation of a novel physician-report tool for assessing barriers to providing care to autistic patients. BMC Health Serv Res. 2021 Aug 26; https://doi.org/10.1186/s12913-021-06842-1

Conference proceedings:

1. Walsh C, Lydon S, Hehir A, O'Connor P. Development and evaluation of a novel caregiver-report tool to assess barriers to physical healthcare for people on the autism spectrum. Paper presented at Health Services Research UK Conference 2021. July 6th 2021; Virtual event

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3. Walsh C, Lydon S, Geoghegan R, Carey C, Creed M, O'Loughlin L, Walsh E, Byrne D, O'Connor, P. Development and preliminary evaluation of a novel physician-report tool for assessing barriers to providing care to autistic patients. Paper presented at Trinity Health

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4. Walsh C, Lydon S, & O'Connor P. Using patient narratives to explore challenging healthcare encounters. Paper presented at Trinity Health and Education International Research Conference 2021; March 9th-11th, 2021. Virtual event.

5. Walsh C, Lydon S, Hehir A, O'Connor P. Development and evaluation of a novel caregiver-report tool to assess barriers to physical healthcare for people on the autism spectrum. Poster presented at Trinity Health and Education International Research Conference 2021; March 9th-11th, 2021. Virtual event.

6. Walsh C, Lydon S, Walsh E, O'Connor P. A systematic review of interventions to improve healthcare experiences and access in autism. Poster presented at Trinity Health and Education International Research Conference 2021; March 9th-11th, 2021. Dublin (Virtual event)

7. Walsh C, Lydon S, O'Dowd E, O'Connor P. Barriers to healthcare for persons on the autism spectrum: A systematic review of the literature and development of a taxonomy. Poster presented at International Forum of Quality and Safety in Healthcare Conference 2020; April 28th-30th, 2020. Virtual event.

8. Walsh C, Lydon S, O'Dowd E, O'Connor P. Barriers to healthcare for persons on the autism spectrum: A systematic review of the literature and development of a taxonomy. Poster presented at National Patient Safety Office Conference 2019; November 13th, 2019; Dublin, Ireland.

9. Walsh C, Lydon S, O'Dowd E, O'Connor P. Barriers to healthcare for persons on the autism spectrum: A systematic review of the literature and development of a taxonomy. Poster presented at National Patient Safety Office Conference 2018; October 17th-18th 2018; Dublin Ireland

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List of Abbreviations

ADHD	Attention Deficit Hyperactivity Disorder
AHAT	Autism Healthcare Accommodations Tool
AIDS	Acquired Immunodeficiency Syndrome
AASPIRE	Academic Autistic Spectrum Partnership in Research and Education
CFA	Confirmatory Factor Analysis
CINAHL	Cumulative Index to Nursing and Allied Health Literature
CIT	Critical Incident Technique
DSM-IV	Diagnostic and Statistical Manual of Mental Disorders 4th edition
DSM-V	Diagnostic and Statistical Manual of Mental Disorders 5h edition
ED	Emergency Department
EFA	Exploratory Factory Analysis
EU	European Union
GAP	General Adjustment Problems
GP	General practitioner
HCAT	Healthcare Complaints Analysis Tool
HCAT-GP	Healthcare Complaints Analysis Tool – General Practice
HCP	Healthcare provider
HIV	Human Immune-Deficiency Virus
HPA	Hypothalamic-Pituitary-Adrenal
HSE	Health Service Executive
ICD	International Classification of Diseases
ID	Intellectual Disability
IDD	Intellectual and Developmental Disabilities
IM	Intervention Mapping
IQ	Intelligence Quotient
KMO	Kaiser-Meyer-Olkin
LGBTQI+	Lesbian, Gay, Bisexual, Transgender, Queer, Intersexed +
MCAR	Missing Completely at Random
MeSH	Medical Subject Headings
MSA	Measures of Sampling Adequacy
MRI	Magnetic Resonance Imaging
NOS	Not otherwise specified
NHS	National Health Service

PA	Parallel Analysis
PCC	Primary care centre
PPI	Public and Patient Involvement
PRISMA	Preferred Reporting Items for Systematic Reviews and Meta-Analyses
PROSPERO	International Prospective Register of Systematic Reviews
QATSDD	Quality Assessment Tool for Studies with Diverse Designs
QI	Quality Improvement
OCD	Obsessive Compulsive Disorder
SHO	Senior House Officer
SP	Standardised Patient
SPSS	Statistical Package for the Social Sciences
SRS-2	Social Responsiveness Scale-2
UK	United Kingdom
UNCRPD	United Nations Convention for the Rights of Persons with Disabilities
USA	United States of America
WHO	World Health Organization

Chapter 1.

General Introduction

Introduction

Improving the physical health and wellbeing of autistic[1] people has been established as a research priority (1, 2, 3). In a workshop co-hosted by Autistica (a UK Autism charity) and the University of Newcastle, 11 priority research concepts related to physical healthcare were identified by autistic individuals, relatives, clinicians, managers, international researchers, and funders. One of these concepts focused on the need for research on 'autism-friendly' healthcare services. Workshop attendees proposed that this should include collating evidence on autism-specific service accommodation and design, establishing best practice guidelines for providing care to autistic individuals, identifying gaps in healthcare provider knowledge, skills, and values, and identifying strategies to change service models to enable a more inclusive health service. These recommendations were supported by a recent systematic review examining the research priorities of the autism community, in which physical health and well-being, and research on developing expertise, coordination, availability and accessibility of services across the life span were identified as leading research priorities in five out of the seven included studies (4).

Clearly, improving physical health and healthcare for autistic individuals is important. Yet, to date, this topic has received less attention than other areas in which autistic people often require supports (education or social skills) or where research is required to advance knowledge of appropriate supports (1, 5). Accordingly, the primary aim of the current thesis is to develop an improved understanding of the barriers to adequate

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[1] I have deliberately opted to use identity-first language (i.e., autistic individual) rather than person-first language (i.e., individual with autism) throughout this thesis. The appropriate use of language around autism is recognised as a complex issue (6) However, I have made this decision as, in recent years, autistic individuals have expressed a strong preference for the use of identity-first language (7,8) and the use of person-first language has been suggested to perpetuate or sustain stigma around disability (9) .

and effective healthcare provision for autistic individuals. This information will facilitate improvement or change within healthcare service provision in order to improve the care of autistic individuals. A secondary aim is to consider how interventions evaluated in research to date might address the barriers to accessing healthcare identified through engagement with autistic individuals, caregivers, and healthcare professionals (HCPs) in Ireland. This first chapter of my PhD will introduce and define autism, consider health disparities and outcomes experienced by the autistic community, and examine the potential determinants of poor health. Subsequently, the theories of healthcare access which have influenced this thesis will be described. This chapter will end with the presentation of an overview of the thesis.

Autism Spectrum Disorder

Autism Spectrum Disorder (hereafter ‘autism’) is a complex lifelong neurodevelopmental condition which is characterised by impairments in social interaction and communication skills, along with restricted, repetitive, and stereotyped patterns of behaviour (10). Although there are core diagnostic criteria for the condition, there is wide heterogeneity in how autism manifests across individuals. The concept of a ‘spectrum’ was first introduced in the 4th edition of the Diagnostic and Statistical Manual (11) in order to account for this heterogeneity in presentation. The DSM-V (10) and the International Classification of Diseases 11 (12) later reclassified autism as a ‘continuous spectrum’, ranging from those who have at least, if not above, average IQ and no history of language delay to those who have no functional language and severe developmental delay (13). In addition to these core diagnostic criteria, autistic individuals often also experience a range of other nondiagnostic conditions and behaviours, including anxiety, depression, sleep and eating disorders, challenging behaviours such as self-injury, and other developmental/intellectual disabilities (14-17). Hyper- or hypo-sensitivities to stimuli in the environment are also common among autistic people and were included as part of the diagnostic criteria in the DSM-V (10, 18). The heterogeneity of experiences and presentation of autistic individuals can create substantive challenges in diagnosing autism and in determining the most appropriate on-going care and support (19).

The need to identify how best to support autistic people is imperative as the prevalence of autism has increased sharply over the past three decades (20). There are a number of potential reasons for this increase including, but not limited to, advances in epidemiological research, increased awareness and understanding of autism, improved diagnostic tools and practices, changing diagnostic criteria, and better distinction between autism symptomatology and that of other conditions such as intellectual disability or attention deficit hyperactive disorder (20-23). Although increases in observed prevalence are consistent, estimating accurate prevalence rates is challenging due to variation in the availability and quality of data from various countries (21). Currently, prevalence is estimated at one in 54 children, and one in 45 adults in the USA (24, 25), while the World Health Organization (WHO) estimates global prevalence rates as approximately 1 in 160 persons (26). Prevalence of Autism in Ireland is currently estimated at between 1-1.5% of school age children (27-29). Such prevalence estimates clearly indicate the need for consideration of the challenges experienced by autistic persons and the supports necessary to address or remediate these challenges.

Given the heterogeneity within the presentation of autism, it is perhaps unsurprising that autistic individuals can experience a variety of challenges which necessitate the development and introduction of evidence-based supports. There has been a substantial amount of research focused on addressing challenges experienced by autistic children, though the experiences of autistic adolescents and adults have received relatively limited attention (30, 31). Key challenges or difficulties experienced by autistic individuals, of all ages, can include: challenging behaviour (e.g., self-injury, aggression) which can have substantial negative impacts on the individual's quality of life by limiting their access to communities, activities, and learning opportunities (32-34); atypicalities in social skills which can lead to negative social experiences (35-37); difficulties in educational settings which can have a negative impact on the individuals' educational attainment, school experiences and well-being (38, 39); difficulties obtaining and maintaining employment which has been associated with poor mental health for autistic individuals (40-43), and challenges related to physical health such as increased morbidity and poor health outcomes (15).

Health inequities for autistic individuals

Poor health outcomes are not a necessary consequence of being on the autism spectrum. Rather, they likely reflect inequities in healthcare whereby people on the autism spectrum have more difficulties accessing quality healthcare than others. Health equity is a complex concept that refers to the absence of systemic disparities in health between social groups who have different levels of underlying social advantage/disadvantage (44). In the landmark report '*Crossing the Quality Chasm: A New Heath System for the 21st Century'*, the Institute of Medicine (45) classified health equity as a core domain of healthcare quality which '*should not differ based on the personal characteristics of patients, such as gender, ethnicity and socioeconomic status'* (45 p6). Inequity in health has thus been defined as '*unjust and avoidable differences in health care access, quality, and outcomes*" (46 p.269; 47). Significant health inequities persist, however, whereby people who are disadvantaged, vulnerable and marginalised in society tend to experience poorer health outcomes compared to those higher up the social ladder (44, 48, 49). People with disabilities are one such group that are consistently reported to experience substantial inequities in health

(50). Common indicators of health inequities include increased morbidity (i.e., the presence of health conditions), mortality (i.e., death), high healthcare service use, and unmet healthcare needs (51). All of which are documented for the autism population.

Physical health and mortality

It has been widely evidenced that autistic individuals experience poorer health outcomes than other populations (15, 52-54). For example, in one large database study in the USA, Croen and colleagues (15) found that all 45 medical conditions assessed (e.g., autoimmune disorders, diabetes, allergies, cardiovascular conditions) were more common among autistic adults compared to age- and sex- matched, non-autistic controls. Other studies have also consistently found that conditions such as obesity; (55-57), immune-related conditions (58), hypertension (59), diabetes (60), and epilepsy (54) were significantly elevated in autistic individuals. Although the autistic community is diverse, these health issues appear consistent regardless of demographic characteristics such as age, gender, or the presence or absence of co-occurring conditions such as intellectual disability (ID; 15, 58, 61). In addition to being at increased risk of experiencing such physical health conditions, autistic individuals appear more likely to experience multiple simultaneous health conditions. For example, Jones et al. (53) found that the autistic adults in their sample had a median of 11 co-occurring conditions (range: 0-33), while Karpur et al. (62) found that 54% of 1,253 autistic children were significantly more likely to have more than four co-occurring conditions compared to 14% of children with other disabilities and 0.3% of children with no disabilities. Perhaps unsurprisingly, given the increased frequency of physical illness among autistic persons, the health status of autistic individuals is consistently rated as ‘poor’ or ‘bad’ by both autistic individuals themselves (63), and their caregivers (64, 65). Additionally, the presence of co-occurring physical health conditions has also been associated with poor ratings of health-related quality of life for both autistic children and adults (66, 67).

In addition to the apparent increased incidence of physical health conditions, research also consistently indicates that autistic individuals experience higher mortality rates compared to the general population (68-73). Indeed, compared to mortality statistics for the general population, mortality has been estimated to be two- (74) to ten-fold (73) higher for autistic individuals. In a population-based case control study of two Swedish health registries, Hirvikoski et al. (70) found that both all-cause (i.e., deaths arising from any cause of death) and cause-specific (i.e., deaths resulting from a specific cause of death such as a specific disease) mortality was elevated for autistic individuals compared to gender, age and county of residence matched non-autistic controls. In Hirvikoski et al.'s study, individuals from the general population died at a mean age of 70.2 years, while autistic individuals died at a mean age of 59.9 years. The authors also found that autistic individuals with no co-occurring intellectual or learning disabilities died an average of 16 years earlier than the general population, and those who had co-occurring ID died an average of 30 years earlier. Elevated mortality risk for those with co-occurring ID has also been observed as particularly high in other studies (e.g., 69, 70, 73) suggesting that although these mortality data are not fully explained by the presence or absence of co-occurring ID, there may be some relationship between these variables. Studies have also consistently found that mortality risk is particularly high for autistic women (e.g., 69-72, 75, 76). Finally, mortality also appears to be elevated within almost all analysed categories of physical health (e.g., diseases of the endocrine system, nervous system, circulatory system; 69-71) suggesting that it cannot be accounted for by the presence of any one physical health condition.

Health service use

A second key indicator of health inequities is health service use. Considering the frequency of co-occurring physical health conditions and elevated risk for mortality in autism, it can be anticipated that autistic children and adults tend to have higher utilisation rates than non-autistic individuals (77-82) across primary (i.e., healthcare provided in the community by professionals such as GPs or community health nurses as a first point of contact with the healthcare services), secondary (i.e., specialised care which is generally provided within hospital settings often by referral from a primary care provider), and emergency care services (i.e., inpatient and outpatient healthcare services that are necessary to prevent serious impairment or death). In a large case-control study (*n*=1,507 autistic adults in the USA), Zerbo et al. (83) found that, even after controlling for demographics and co-occurring medical and psychiatric conditions, autistic adults had a significantly higher number of primary care visits than non-autistic adults with Attention Deficit Hyperactivity Disorder (ADHD), and adults from the general population. This finding of higher usage of primary care services among autistic individuals compared to non-autistic individuals is consistent in the research, even when controlling for demographic characteristics such as age, sex, and ethnicity (81, 84) or co-occurring physical and mental health conditions (85). These results are also consistent across the studies of primary care use among both autistic children (85) and adults (86). Importantly, higher usage of primary care services does not appear to result in improved health or quality of care for autistic individuals. Despite the higher rates of primary care contact, autistic individuals tend to have lower rates of preventative care, including vaccinations (79, 85, 82), well-child visits (85), gynaecological visits (83), and cancer screening (79, 83). Additionally, caregivers and autistic individuals often indicate low satisfaction with their primary care providers (79, 82). Improving access to good quality primary and preventative care is crucial as better access to primary care has been associated with reductions in hospital admissions and emergency care usage (87-89).

Research has also indicated that autistic individuals tend to have more frequent contact with secondary healthcare services (52, 82, 85). Croen et al. (90) found that autistic children had approximately 40% more outpatient visits to paediatrics and neurology than non-autistic children. These findings were echoed by Liptak et al. (82) who also found that autistic children had significantly more annual out-patient and physician visits than children from the general population. In addition to attendance at secondary healthcare services, hospitalisation rates have also been found to be higher for autistic individuals compared to the general population (90, 91), as well as compared to individuals with ID and psychiatric conditions (92). Croen et al. (90) for example, found that autistic children experienced significantly more inpatient (3% vs 1%) and outpatient (5% vs 2%) hospitalisations compared to non-autistic children. Further, in addition to increased hospitalisation rates, autistic individuals also tend to have longer hospital stays than the general population (81, 93). Mandell et al. (92) found that autistic children had a mean stay of 25 days, compared to children with intellectual disabilities who had a mean stay of 13 days, and children with other developmental or psychiatric conditions who had a mean stay of five days. While Croen et al. (90) found that autistic children had almost four times more inpatient hospital days compared to non-autistic children. These findings of higher rates of contact with secondary services by autistic individuals appear to be consistent across the autism population, regardless of factors such as age, gender or the presence or absence of co-occurring conditions (85, 90, 94, 95). However, these higher utilisation rates do not appear to be associated with better outcomes as numerous studies have found that caregivers and autistic individuals are often unsatisfied with hospital care (96-99) and Akobirshoev et al. (76) found that autistic individuals experience higher inpatient mortality than non-autistic controls.

Use of the emergency care services, which includes the emergency department (ED), is also high among autistic individuals (79, 90, 100, 101). In a recent analysis of emergency department visits in all hospitals across the state of New York, Beverly et al. (102) found that, on average, autistic children, and adolescents between the ages of 2 and 22 years had significantly more ED visits than non-autistic children and adolescents and were significantly more likely to exhibit frequent ED use (i.e., more than four ED visits per year; 102-104). Similar to rates of utilisation of other healthcare services, high ED use appears to be experienced by the entire autism population regardless of factors such as age, gender or the presence or absence of co-occurring conditions (52, 79, 86, 105). Importantly, although research has indicated that autistic individuals are often more likely than non-autistic individuals to be hospitalised as a result of an ED visit (81, 86, 95, 102), a number of studies have found that many visits to the ED by autistic individuals are for non-urgent reasons suggesting that there may be barriers in place which prevent autistic individuals getting the care they need in non-emergency settings (89, 100). Lin et al. (63), for example, found that children with developmental disabilities, including autism, who had higher ED utilisation rates, did not have primary care providers who listened to parents' concerns and did not have strong family partnerships with their primary care provider. In a later study, Lindley et al. (89) found that delays in accessing needed care was the most commonly cited reason for presenting to the ED as opposed to a community healthcare setting. Other reasons included not finding a primary care provider who would take the autistic child as a patient, and not having a primary care centre nearby (89). Thus, it appears that at least some of the high usage of ED by autistic individuals can be accounted for by the presence of unmet needs in other services.

Unmet needs

Research has consistently shown that despite high rates of contact with the healthcare services, autistic individuals are more likely to experience substantial unmet healthcare needs across a range of services relating to physical health. Unmet healthcare needs, which are another indicator of healthcare inequities, are defined as '*the difference, if any, between services that are judged necessary to deal appropriately with health problems and the services actually received…an unmet need is the absence of any, or of sufficient, or of appropriate care and services*' (106 p.418). Some examples experienced by the autism community include unmet needs for family-centred, comprehensive, or coordinated care (107), services to support transition from paediatric to adult healthcare services (66), and preventative and specialty care (78). Utilising data from the 2016 National Survey of Children's Health in the USA, Karpur et al. (62) found that autistic children were 15 times more likely to have unmet healthcare needs as compared to children without disabilities. Almost 16% of autistic children did not receive family-centred care, compared to 11% of children with other disabilities, and 8% of children with no disabilities, even when controlling for predisposing factors (e.g., age, gender), enabling factors (e.g., access to health insurance), and need-based factors (e.g., co-occurring conditions). Data on higher unmet need among autistic individuals have also been consistently observed when comparing data from autistic individuals to that taken from other populations including ID and psychiatric conditions (77, 78).

Although comparatively little research has examined unmet needs for autistic adults, similar patterns are apparent in studies (79, 94). Nicolaidis et al. (79) found that 30% of autistic adults (*n*=209) reported unmet needs related to physical health problems, preventative care, and prescription medications compared to 16% of non-autistic adults (*n*=228). Thus, the research indicates that unmet healthcare needs and the other core indicators of health inequity (i.e., increased morbidity and mortality, high healthcare utilisation) have been clearly, and consistently, documented among autistic persons. In this way, health inequity has significant impacts for the autistic community. In order to reduce health inequity for autistic individuals, it is necessary to first consider, and develop an understanding of the potential determinants of this inequity so that targeted action may be taken to address the issue.

Determinants of health inequities for autistic individuals

A number of determinants of health inequity have been suggested (47). The most common of which include genetic/biological factors (e.g., age, genetic predispositions to certain conditions) and socio-economic factors (e.g., income and social protection, housing and living conditions, food insecurity, employment status, working life conditions, early childhood development, social inclusion, education, access to quality healthcare (48, 108). Health differences that are attributable to natural biological variation, such as those that arise as a result of the aging process, can be considered inevitable and tend not to be considered inequitable (i.e., unjust or unfair; 47). However, many socio-economic determinants of health are not inevitable. For example, people of lower socio-economic status often suffer poorer health as a result of poor living and working conditions in which they are forced to remain due to a lack of resources (47). Many of these social determinants are amenable to intervention and addressing those determinants could have a positive impact on health outcomes. For example, in a systematic review, Black et al. (109) found evidence to suggest that participating in food subsidy programmes can have positive impacts on health equity and health status for disadvantaged groups. Specifically, evaluated subsidy programmes were found to increase fruit and vegetable intake in women; improve the nutritional status of pregnant women; and increase mean birthweights of infants. Similarly, Thomson et al. (110) found, in a systematic review, that interventions aimed at improving housing, such as refurbishment and improving energy efficiency, were associated with improved health outcomes (e.g., improved respiratory symptoms, reduced illness episodes), and reduced healthcare service use (e.g., less GP visits, reduced likelihood of inpatient/outpatient service use). Little research has probed the determinants of health inequity specific to autism. However, there has been a relatively robust investigation of health inequity among persons with ID. Emerson and Baines (11) highlighted a number of determinants of health inequities for people with ID, including: 1) genetic/biological factors; 2) communication and health literacy; 3) social determinants; 4) personal health risks and behaviours; and 5) deficiencies in access to quality healthcare. It is reasonable to suggest that such factors play a role in perpetuating health inequities for autistic individuals. For instance, research shows that autistic individuals are more likely to be less educated, unmarried, and more socially deprived than the general population, and tend to have difficulties accessing quality healthcare (40, 112-114).

Genetic and biological factors

The genetic or biological basis of certain IDs may be a risk factor for certain physical or mental health conditions which negatively impact health (115). For example, people with Down’s syndrome have a high risk of congenital heart defects (116, 117). Biological vulnerability may also play a role in adverse health outcomes in autism. For example, shortened telomere length in peripheral blood leukocytes which are associated with a variety of physical health (e.g., cancer, cardiovascular diseases; 118, 119) and psychiatric conditions (e.g., schizophrenia; anxiety disorders; 120, 121), has also been found to be associated with autism (122). Further, dysregulation of the hypothalamic-pituitary-adrenal (HPA) system, which is associated with adverse effects of the immune system (123), has been commonly observed among autistic individuals who experience more severe cognitive impairment (124). These data, along with the high rate of co-occurrence of some health conditions indicate that more research is required on potential biological or genetic links between autism and certain health conditions in order to understand whether clear risk factors exist.

Communication and health literacy

Communication difficulties, which are part of the diagnostic criteria for autism, create challenges in healthcare settings by impeding patient-HCP communication. For example, due to slower language processing speeds and anxiety caused by healthcare settings, some autistic individuals may have difficulties keeping pace with the conversation during a typical consultation and so may miss important medical information or the opportunity to ask questions (98, 113). In addition, some autistic individuals report difficulties communicating their symptoms to healthcare professionals (22), while caregivers and healthcare providers often have difficulty interpreting the autistic person's behaviours or symptoms, especially if the autistic person cannot communicate verbally (113, 125). Such communication difficulties can lead to diagnostic overshadowing or missed diagnoses which can have substantial adverse effects on health (126, 127).

Another factor that contributes to health communication is health literacy, which has been defined as '*the degree to which individuals can obtain, process, understand, and communicate about health related information needed to make informed health decisions*' (128, p.16; 129). Research has consistently reported that people with intellectual disabilities and/or autism often find health information incomprehensible for various reasons including the HCP not using, or health information not being presented in, accessible language (97, 113, 130). Inaccessible information is a contributing factor to low levels of health literacy, which have been associated with a wide range of adverse impacts on the care process and health outcomes including higher health service use, lower rates of preventative care, poor health behaviours, and poorer overall health status (128, 131, 132). It is therefore essential that efforts are made to promote and enhance health literacy for people with ID and/or autism. Practical solutions such as communication training for HCPs or developing accessible resources for health-related information (e.g., easy-to-read versions of information leaflets) may help to address this determinant of health inequity and so warrant further investigation with autism populations (133-136).

Social determinants

A number of social and economic factors are widely recognised to impact on health. Social determinants of health refer to '*the conditions in which people are born, grow, work, live and age and the systems put in place to deal with illness* (48, p.xvi; 137, 38). Factors that contribute to social determinants of health include income, housing, food insecurity, employment status, working life conditions, social inclusion, and education (47, 48, 111, 139). Research has indicated that autistic individuals are prone to experiencing many of these social determinants of health including high rates of unemployment (43, 140), social exclusion and loneliness (141-143) discrimination and stigma (130, 144), and poverty (145, 146). In addition, certain subgroups within the autism population including individuals who are members of ethnic minorities or the LGBTQI+ community are at even greater risk of experiencing many of these social determinants of health (147, 148). As social determinants have been cited as particularly important in relation to health (48, 138), it is imperative that research focus on reducing the exposure of autistic individuals to common social determinants in order to improve health outcomes. In recent years, some attention has been given to improving the employment rates of autistic individuals (43, 149). Developing and implementing interventions aimed at improving employment rates for this population may improve income, which may help avoid the downward social mobility often experienced by people with disabilities, and thus improve health outcomes (47, 150). Employment is just one social determinant, however, and although the various social determinants may interact with one another, it is important to consider each in their own right to truly improve health equity (47, 48).

Personal health risks and behaviours

Personal health risks and behaviours are factors that can impact health such as smoking, diet, physical activity levels, risky sexual behaviour or substance abuse and research has indicated that many of these factors are implicated for autistic individuals (111). Food aversion and food selectivity, which refer to limiting behaviours in relation to food and eating, are often exhibited by autistic individuals and can be a product of sensory issues or restricted behaviours or interests (10, 151-153). As a result, food aversion or selectivity can lead to poor nutrition, underweight, or overweight due to restricted or unbalanced diets (154-156). An additional risk factor for overweight is insufficient amounts of daily physical activity which has also been documented as a common issue for many autistic individuals (157-159). A sizable amount of research exists which targets diet and physical activity behaviours for autistic people (160). However, there is less focus on some of the other important health behaviours, such as those relating to sexual health. Research indicates that autistic adolescents experience heightened sexual health risks (161, 162), yet evidence suggests that autistic adolescents face barriers to accessing sexual health services and the informal channels (e.g., peers) through which young people usually learn about sex (58, 163, 164). In addition, research has indicated that HCPs and caregivers are often reluctant or unsure of how to discuss issues around sex with autistic individuals (58, 165). Thus, researchers have consistently recommended developing caregiver and HCP training to facilitate communication and education about sexuality and sexual health for autistic individuals as one means of addressing this determinant of health (166, 167).

Poor access to quality healthcare

Finally, poor access to quality healthcare can also have substantial negative impacts on health. According to the Institute of Medicine (45), quality healthcare is: 1) *safe* (i.e., avoids harming patients from care that is intended to help them); 2) *effective* (i.e., provides evidence-based services to all who could benefit from them and refrains from providing services to those who are not likely to benefit from them); 3*) person-centred* (i.e., is respectful of and responsive to individual patient preferences, needs and values and ensures that all clinical decisions are guided by patient values); 4) *timely* (reduces waits and harmful delays for both those who give and those who receive care); 5) *efficient* (i.e., avoids waste, including waste of equipment, supplies, energy and ideas); and 6) *equitable* (i.e., does not vary in quality because of personal characteristics such as gender, ethnicity, geographic location and socioeconomic status). It is well documented that access can vary according to personal characteristics resulting in some individuals receiving poorer quality care than others and this has been suggested to be significant problem within autism (77, 78, 107, 113, 168). Considering these data, it is perhaps unsurprising that autistic individuals and their caregivers often report feeling dissatisfied with the quality of care they receive (113, 169). Improving access to care has been associated with improved satisfaction with the care received, however, which further elucidates the need for research focused on improving access to care for the autism population (170).

Access to healthcare: definitions and theories

Access to healthcare is a complex but important concern for health policy and the performance of healthcare systems internationally. Consequently, it has been considered extensively in health services research, with much of the focus of this research relating to patient satisfaction and patient utilisation of services (171-173). In the 1960s and 1970s, healthcare utilisation theories dominated health services research and although ‘access’ was viewed as an important concept or determinant of utilisation within many of these theories, access as a specific concept often remained ambiguous and unmeasured (e.g., 174, 175). Subsequently, interest began to shift away from ‘utilisation’ and towards ‘access’ as a concept in its own right. This shift is as a result of repeated observations that mere entry into, or use of, healthcare services could not be fully explained by the health status or health concerns of patients (171, 176). Viewing access as ‘use’ suggests that individuals who do not use services, or use services differently, experience differences in access, however, this is not necessarily the case as, for example, some individuals choose not to use services based on religious or cultural beliefs (177). Thus, access theories began to focus more on the characteristics of healthcare system (i.e., supply factors) rather than the patients’ health seeking behaviours (i.e., demand side factors; 178). Access as a concept is not easily defined, however, and in the years since, numerous theories and definitions of access have been conceptualised. These theories and definitions have been manifold and have differed substantively.

Informed by the determinants of utilisation proposed by earlier utilisation theorists (e.g., 174, 175), Penchansky and Thomas (171) were the first to develop a ‘theory of access’ which provided a useful definition of access. In their model, access was conceptualised as the degree of ‘fit’ between the characteristics of health services and providers, and the characteristics of healthcare consumers; in essence, access is about whether the healthcare service is appropriate to the needs of patients. The framework describes five distinct dimensions of access: (1) *Availability,* which relates to the supply and demand of the healthcare services. Specifically, the relationship between the extent to which the HCP has the required resources to adequately meet the clients' needs; (2) *Accessibility*, which is determined by the geographic accessibility of the services relative to the client and encompasses factors such as transportation resources, travel time, distance and cost; (3) *Accommodation*, which is determined by the extent to which services are organised in a way that can meet the preferences and constraints of the client (e.g., how telephone services are organised and operated, the potential for walk-in appointments, and the opening hours of a facility); (4) *Affordability,* which is determined by how the HCPs prices/costs relate to the clients income, ability or willingness to pay, and health insurance coverage. Important to this domain is client's perception of the worth of the service relative to the cost, and the client's knowledge of cost-related factors; and (5) *Acceptability* which is determined by how comfortable the consumers are with characteristics of the HCP (e.g., gender, age, ethnicity, location of healthcare facility), as well as the HCPs attitudes/preferences regarding acceptable characteristics of consumers. For example, HCPs may be less willing to see clients who are in receipt of welfare payments or who have certain diagnoses. Within this theory, each dimension is important in the assessment of access; addressing one dimension alone is considered unlikely to improve access if problems still exist with the other dimensions. For example, improving the availability or supply of services is unlikely to improve access if affordability barriers continue to exist (179). This theory remains one of the most influential theories of healthcare access, and has formed the basis of many subsequent theories (e.g., 176, 180).

Frenk (180) later expanded on Penchansky and Thomas's (171) theory and suggested that 'fit' is a process of adjustment between the healthcare system and the population. According to Frenk (180), the terms access and accessibility must be defined separately: ‘access’ (also referred to as ‘utilisation power’) is the ability of the population to seek and obtain care, while ‘accessibility’ is the degree of adjustment between the characteristics of the health care resources and those of the population who are in the process of seeking care. Thus, in this theory, access is purely the use of service and accessibility relates to the experience of use and suitability of care. Important to this framework are the concepts of: (1) *availability* (i.e., the existence of healthcare resources and their capacity to produce services) and (2) *resistance* (i.e., the obstacles that arise from healthcare resources that prevent consumers from seeking and obtaining care, including the cost, location, and organisation of services). This consideration of obstacles or barriers to access as a mediator between availability and the use of services became an important consideration in a number of subsequent theories (e.g., 172, 181).

Next, Margolis et al. (181) defined access as the timely use of personal health services to achieve the best possible outcomes. Thus, in this theory, access refers to getting healthcare that is needed when it is needed. This theory described three key dimensions of access: (1) *The Structural dimension* relates to the availability of HCPs and non-fragmented care, as well as the organisation of services (i.e., difficulties making appointments, opening hours of facilities, waiting times); (2) *the personal dimension* relates to patients’ knowledge and attitudes about health and health services, capacity to seek and obtain care, acceptability of services, cultural and language influences; (3) *the financial dimension* relates to insurance coverage, reimbursement rates, and public support. The model also suggests that the appropriateness and efficacy of treatment, the quality of healthcare providers, and adherence to treatment act as mediators between the use of services and health outcomes. In application of the model, Margolis et al. (181) place higher emphasis on targeting the structural and personal barriers for improving access to care and as a result, this model has been referenced within research on access to healthcare for socially disadvantaged children in order to highlight that there are barriers other than financial barriers for this population (182, 183).

Next, Shengelia et al. (178) defined access in terms of effective coverage (i.e., the likelihood of receiving a necessary health intervention when it is needed), and utilisation (i.e., the quantity of healthcare services and procedures used). Thus, in this theory access refers to possibility of obtaining and using needed healthcare when it is needed. This theory also incorporates a number of dimensions: (1) *physical accessibility* (i.e., the extent to which a health intervention is physically accessible to the population, taking into account factors such as time and distance); (2) *resource availability* (i.e., the extent to which sufficient amounts of resources and technologies are available to deliver a health intervention); (3) *cultural acceptability* (i.e., the extent to which services are culturally acceptable to the population); (4) *affordability* (i.e., the amount of an individual's disposable income for health and how healthcare finance is organised within a country); (5) *provider-related quality* (i.e., the ability of providers to use the available technologies and resources for producing health gains for consumers); (6) *adherence* (the ability of consumers to adhere to the treatment regimen); and (7) *strategic choice* (i.e., the possibility of choosing the most effective intervention from potential choices for a given health condition). This theory differed slightly from other theories as the authors suggest that the provision of services can be measured more comprehensively by evaluating the 'effective coverage' of the healthcare system. The authors argue that effective coverage helps to clarify the interactions between the concepts of access, demand, utilisation and coverage'. This model has been used in assessments of the level of health system coverage in Mexico (184, 185).

Peters et al. (186) also defined access as the actual use of services with a clear emphasis on the characteristics of both users and services.

Similar to Penchansky &Thomas (171), Peters et al., (186) also defined access as the appropriateness of the health services to the needs of the patients. Peters et al.'s (186) model has four main dimensions: (1) *geographic accessibility* (i.e., the physical distance from, and travel time to, the healthcare facility); (2) *availability* (i.e., that the right type of care is available to those who need it, encompassing hours of operation, waiting times, appropriate service provider and materials); (3) *Financial accessibility* (i.e., the relationship between the price of services and the consumers' willingness and ability to pay for the services, as well as the level of protection from economic consequences of health costs); (4) *Acceptability* (i.e., the fit between how responsive the service providers are to the social and cultural expectations of community and individuals users). This model has been widely applied to assess disparities in health in developing countries (187-189).

McIntyre et al. (177), defined access as empowerment. Thus, in this theory, access is determined by the level of 'fit' between the services and the individual's ability to use the services. Similar to earlier models (e.g., 171, 180), McIntrye et al. (177) also consider the interaction between supply and demand side factors is important. This model comprises three dimensions: (1) *Availability* which refers to whether appropriate services and provides are available in the right place, at the right time to meet the needs of the population. Important factors within this construct include the interactions between the location of the services and the location and transport options of the population, or the ability and willingness of the services and providers to provide care in accordance with the type and severity of the health conditions of the population; (2) *Affordability* refers to degree of fit between the full cost of the service and the population's ability and willingness to pay for the service and includes factors such as the price of services, public funding levels, other direct costs such as transport, and indirect costs such as lost income. This construct also takes into consideration the impact of cost on household wellbeing when using household resources to cover costs; (3) *Acceptability* refers to the fit between the providers and patients' attitudes towards each other based on certain characteristics (e.g., HCPs may be less accommodating to patients with certain health conditions, or patients may be less willing to receive care from a HCP of a certain ethnicity). Although the dimensions are distinct constructs, they also interact with one another to influence access, for example, *availability* of only male HCPs can create *acceptability* problems for female patients who are only comfortable with female HCPs. This consideration of access as empowerment appears to have marked a shift toward more patient-centred conceptualisations of access.

More recently, Levesque et al. (172) also aimed to develop a more patient-centred conceptualisation of access, incorporating both the supply-side (characteristics of the services) and the demand-side (characteristics of consumers) of access. In this framework, access is defined as '*the opportunity to reach and obtain appropriate healthcare services in situations of perceived need for care'* (172, p. 4). Thus, in this theory access refers to the interaction between the characteristics of the population and the characteristics of the system. In this framework, access is operationalised by distinguishing the various steps throughout the entire process of care and comprises five supply-side dimensions which build on previously proposed dimensions (e.g., 171, 178, 180, 186): (1) *Approachability*, which is determined by the capability of people with healthcare needs to identify that services exist, can be reached and have an impact on the health of the individual; (2) *Acceptability*, which is determined by social and cultural factors influencing the possibility for people to accept characteristics of the services (e.g., sex or social group of the HCPs) and the judged appropriateness of persons to seek care (e.g., religious beliefs); (3) *Availability and Accommodation*, which are determined by whether services can be reached physically and in a timely manner, encompassing factors such as geography and opening hours; (4) *Affordability* which is determined by the economic capacity of people to spend resources and time to use appropriate services; and (5) *Appropriateness* which is determined by the fit between the services and the client's need, the timeliness of care, the amount of time spent in assessing health problems and determining the treatment plans ,and the technical and interpersonal quality of the services provided. The authors note that barriers to care can occur within each of these dimensions and so, gaining access requires that the population possess five corresponding abilities to overcome potential barriers: (1) the ability to perceive a need for care; (2) the ability to seek care; (3) the ability to reach care; (4) the ability to pay for care; and (5) the ability to engage with care. The constructs within the framework are interdependent and are arranged in pairs – each supply-side dimension of accessibility is mirrored by a matching demand-side ability. Similar to McIntyre et al. (177), the framework emphasises that it is the interactions between the supply and demand side factors that determine access, for example, is not just the location of the healthcare facility that determines access, rather it is the interaction between the location and the patient's ability to travel there. This person-centred conceptualisation is slightly different from earlier models which placed more emphasis on the supply-side dimensions. As a result, this model has been widely applied to assessments of access to healthcare for a variety of marginalised populations including LGBTQI+, ethnic and indigenous communities (e.g., 190-192).

Finally, Saurman (176) describes an expansion of Penchansky and Thomas's (171) 'fit' theory to include the dimension of '*Awareness*', which the author reports became apparent in an earlier study in which a mental health emergency access programme in rural Australia was evaluated (193). The authors found that many participants were unaware of the programme, indicating that awareness was an important component of access (193). Awareness is viewed as bi-directional: services that are aware of the consumers' needs can provide more effective care for that population, whereas in order to access a service, consumers need to be aware that a service exists. The dimension of Awareness in Saurman's theory encompasses the components: (1) *communication*, which is achieved by establishing sustainable and targeted communication strategies to raise and maintain consumer awareness; and (2) *health literacy* which is the output of effective communication and information sharing. Health literacy, in this framework, involves accessing, understanding, and using information to make health decisions and is critical to the empowerment of both patients and HCPs. Saurman argues that the addition of awareness to Penchanky and Thomas' (171) theory strengthens the conceptual framework, providing a more comprehensive conceptualisation of access.

The described models and frameworks of access include various dimensions, some of which are similar across models, some which use similar labels for different meanings, and some are modifications of previous dimensions. For example, Penchansky & Thomas's (171) dimension of accessibility has been split into financial and geographic accessibility by Peters et al. (186). This demonstrates the complexity of conceptualising, defining, and measuring access to healthcare. Regardless, there is a general thread across frameworks and definitions that access is a multi-dimensional concept which relates to the ability of an individual with a healthcare need to obtain the right care from the right provider, at the right time, in the right place, dependent on context (176). It is also clear from these theories that barriers to access can stem from both the supply-side (i.e., the healthcare service or the provider) and demand-side (i.e., the patient), and that these can interact, meaning that both aspects need to be considered in any evaluation of access (194, 195). This also suggests that factors which influence access to healthcare, and therefore physical health, are amenable at the level of the system (changing the way healthcare systems function; expanding the scope of practice) and at the individual or population level (e.g., empowering patients to engage in shared decision making).

Access to care is now well recognised as key to supporting the good health of populations, thus, there is a clear need to examine access, and the different aspects (e.g., accommodation, affordability, availability) that have been documented across these various theories of access over the past decades. Developing an understanding of access to healthcare for autistic persons will be key to elucidating factors which may contribute to the poor physical health outcomes, excess mortality, and unmet healthcare needs among the autism population. Engagement with all relevant stakeholders (i.e., autistic individuals, the caregivers of autistic individuals, and HCPs) is required to develop this understanding, but doing so requires possession of the appropriate tools to evaluate or elicit an understanding of access and the factors that impede it. Developing such an understanding would support targeted interventions to improve access to healthcare for autistic individuals which may translate to an improved quality of care and better health outcomes for individuals on the autism spectrum.

Overview of the thesis and research questions

In order to improve access to healthcare, a thorough understanding of the experiences of barriers to, and quality of, healthcare for people on the autism spectrum is essential. This understanding will require engaging with all relevant stakeholders, including autistic individuals, caregivers, and HCPs to gather both international and national perspectives of barriers to care for the autism population. In order to gain a holistic view, a variety of research methods are required, including systematic assessment of international literature, qualitative examinations, and valid and reliable measurement tools to assess barriers to care. This would support the development of effective interventions and quality improvement initiatives to improve access to and the delivery of healthcare for the autism population.

The central aim of the proposed research is to examine the barriers to accessing care for autistic individuals and consider how access might be improved. This thesis is centred around five research questions, each addressed in a separate chapter. Chapter 2 will address the question: *what are the barriers to healthcare access that have been reported internationally by autistic individuals, caregivers, and HCPs?* Understanding the barriers to healthcare access that have been reported internationally by various stakeholders will allow comparisons to be drawn between groups and help to identify the types of interventions and resources which are most required. To date, no systematic review has collated the extant research on barriers to physical healthcare from the perspectives of autistic individuals, caregivers, and HCPs. Thus, Chapter 2 will describe a systematic review of the extant research on barriers to healthcare for people on the autism spectrum.

Chapter 3 will address the question: *what are the barriers to physical healthcare reported by caregivers of autistic individuals living in Ireland?* Much of the research on healthcare and autism has relied on caregiver-report. However, there is a lack of caregiver-report measurement tools which can assess barriers to healthcare which occur across the healthcare system, and that are informed by barriers identified by autistic individuals. Therefore, the aim of this study was to develop a caregiver-report tool of barriers to care for autistic individuals and to administer this to caregivers of autistic individuals living in Ireland. The final aim of this study was to identify factors which may be associated with the barriers experienced.

Chapter 4 will address the question: *what are the barriers to providing care to people on the autism spectrum experienced by physicians in Ireland?* Despite widespread agreement that HCP knowledge and self-efficacy with regards to caring for autistic individuals is low (196), the findings from the systematic review in Chapter 2 indicated that no existing measurement tool which assessed barriers to care that exist across the healthcare system for physicians has been previously developed. The aim of this second study, thus, was to develop and evaluate a physician-report tool to assess barriers to providing care to people on the autism spectrum and administer this tool to physicians working in Ireland. A final aim was to assess factors which may be associated with those barriers.

Chapter 5 addresses the question: *what barriers contribute to challenging healthcare encounters for people on the autism spectrum?* This third study aimed to use patient narratives to qualitatively explore challenging healthcare encounters involving people on the autism spectrum. Qualitative research is needed to provide context in order to explain how and why barriers to healthcare manifest for individuals on the autism spectrum and to elucidate the impact that barriers have on care experiences and health.

Chapter 6 addresses the question: *what interventions have been implemented to improve experiences of, or access to, healthcare for people on the autism spectrum and how are they evaluated?* There is an urgent need to develop quality improvement initiatives and interventions to actually improve care for autistic individuals. Currently, no systematic review has assessed the evidence for interventions that have been trialled to date with the aim of improving access to or experiences within healthcare. Chapter 6, therefore, describes a systematic review of interventions in order to provide guidance and recommendations for improvement strategies.

Finally, Chapter 7 presents an overall discussion of the research findings in the context of existing literature, including how they relate to contemporary access to healthcare theories. The strengths and limitations of the thesis are considered and recommendations for future research, policy and practice will be discussed. The thesis presented herein comprises a collection of individual studies conducted by the researcher to fulfil a thesis by publication requirement.

Conclusion

Improving physical health and the quality of healthcare for people on the autism spectrum has been consistently identified as a research priority by the autism community (1, 3) and this priority is supported by various international organisations (197-199). This recognition emphasises the importance of examining access to healthcare for this population and underscores the timeliness of this current programme of research. As discussed above, autistic individuals have increased prevalence of many health conditions, experience poorer health outcomes, more unmet healthcare needs, and higher premature mortality rates compared to the general population, despite higher rates of contact with the health services (15, 70). A key contributor is likely to be difficulties experienced by autistic individuals when accessing appropriate and effective healthcare services. Thus, this programme of research will assess the barriers to healthcare for autistic individuals from the perspectives of various stakeholders and consider the next steps in terms of overcoming these barriers.

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Chapter 2: Study 1

Barriers to Healthcare for People on the Autism Spectrum: A Systematic Review of the Literature and Development of a Taxonomy

Barriers to healthcare for people on the autism spectrum: A systematic review of the literature and development of a taxonomy

Declaration

Where this fits in with the thesis

People on the autism spectrum experience substantial health inequities (1, 2, 3) reflected by poorer health outcomes in this population (4, 5). In order to address these inequities, we first need to gain a thorough understanding of the barriers that prevent people from accessing quality healthcare. To date, no systematic review has collated the extant research on barriers to physical healthcare from the perspectives of autistic individuals, caregivers, and healthcare providers. Therefore, Chapter 2 aimed to address the research question: *what are the barriers to healthcare access that have been reported internationally by autistic individuals, caregivers, and HCPs?* The purpose of conducting this review was to provide a basis on which researchers, policy makers and practitioners could develop measurement tools and interventions to assess and overcome barriers to care experienced by the autism population.

Peer-reviewed publication

This study has been accepted and published in a peer-reviewed journal.

The citation is:
Walsh C, Lydon S, O’Dowd E, O’Connor P. Barriers to healthcare for persons with autism: A systematic review of the literature and development of a taxonomy. Developmental Neurorehabilitation. 2020 Oct 2;23(7):413-30.

The following chapter is a formatted version of the submitted manuscript to the journal.

Conference presentations

Oral presentation

Walsh C, Lydon S, O’Dowd E, O’Connor P. Barriers to healthcare for persons with autism: A systematic review of the literature and development of a taxonomy. Paper presented at Trinity Health and Education International Research Conference 2021; March 9th-11th, 2021. Virtual event.

Poster presentations

Walsh C, Lydon S, O’Dowd E, O’Connor P. Barriers to healthcare for persons with autism: A systematic review of the literature and development of a taxonomy. Poster presented at the International Forum of Quality and Safety in Healthcare Conference 2020; April 28th-30th, 2020. Virtual event.

Walsh C, Lydon S, O’Dowd E, O’Connor P. Barriers to healthcare for persons with autism: A systematic review of the literature and development of a taxonomy. Poster presented at the National Patient Safety Office Conference 2019; November 13th, 2019; Dublin, Ireland.

Walsh C, Lydon S, O’Dowd E, O’Connor P. Barriers to healthcare for persons with autism: A systematic review of the literature and development of a taxonomy. Poster presented at the National Patient Safety Office Conference 2018; October 17th-18th 2018; Dublin Ireland

Author contributions

CW, POC and SL proposed and designed the study. CW developed the search strategy, executed the database searches, and conducted the database screening. CW and EOD conducted the data extraction and quality appraisal. CW analysed and synthesised the data. CW drafted the manuscript. CW, SL, and POC contributed to the manuscript. All authors have read, commented on, and approved the final manuscript.

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Abstract

Purpose: This review aimed to: 1) synthesise extant research on barriers to healthcare access experienced by autistic individuals, their caregivers, and healthcare providers; and 2) present a taxonomy of barriers to physical healthcare for autistic individuals.

Method: Systematic searches were conducted in five electronic databases. Methodological rigour was assessed using the Quality Assessment Tool for Studies with Diverse Designs. Thematic analysis was used to classify barriers and to develop a taxonomy.

Results: In total, 31 articles were included in the review. The resulting taxonomy consisted of four themes: 1) Challenges Associated with Autism-related Characteristics; 2) Healthcare Provider-related Issues; 3) Healthcare System-related Issues; and 4) Other Patient-related Factors.

Conclusions: Barriers to healthcare access for autistic individuals are prevalent and occur at the patient, provider, and system levels. The taxonomy developed may facilitate measurement of barriers within healthcare facilities and prompt identification of areas where interventions are warranted to improve care.

Keywords: Autism Spectrum Disorder; Healthcare Access; Barriers; Systematic Review; Health; Reasonable Adjustments.

Conflicts of interest

The authors have no conflicts of interest

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Introduction

Autism is a neurodevelopmental condition characterised by persistent deficits in social and communication skills and repetitive or restricted behaviours and interests (1). Although international prevalence rates vary, current estimates suggest that approximately one in every 160 children worldwide is autistic (2). Autistic individuals are more likely than non-autistic individuals to experience a variety of co-occurring conditions such as gastrointestinal problems or cardiovascular issues (3,4), neurological conditions including epilepsy, schizophrenia, and sleep disorders (5,6), and mental health and psychiatric conditions, such as depression or anxiety (7,8).

The presence of these additional medical comorbidities may be one reason for the higher frequency with which autistic individuals typically engage with healthcare systems compared to the general population. Research shows that autistic individuals have a greater number of physician visits per year and attend for both non-emergency and emergency hospital care with more regularity (3,9). However, research has also shown that autistic individuals have lower engagement with some preventative healthcare services. For example, Nicolaidis and colleagues (10) reported that only 59% of autistic women in their study had received a pap smear in the past three years as compared to 78% of non-autistic controls. Similarly, Liptak et al. (11) found that autistic children were less likely to have injections provided or ordered during a healthcare visit compared to children with other special healthcare needs. Both autistic children and adults have also been found to have a higher incidence of unmet healthcare needs as compared to typically developing persons (10,12). Such unmet healthcare needs may relate to the poorer health outcomes (7,13), lower health related quality of life (14,15), and increased mortality (16,17) that has been observed among autistic individuals compared to the general population.

These data are suggestive of a disparity in access to appropriate healthcare services among autistic individuals and emphasise the necessity of examining the functioning of healthcare services, along with exploring the experience of autistic patients, their caregivers and the clinicians that care for them in healthcare settings (10,18). Healthcare access has been defined in a variety of ways. Penchansky and Thomas (19, p.128), for example, define healthcare access as a wider concept ‘representing the degree of ‘fit’ between the clients and the system’, encompassing the factors of availability, accessibility, accommodation, affordability, and acceptability. Similarly, Shengelia et al. (20) define access in terms of effective coverage (i.e., the likelihood of receiving a necessary health intervention when it is needed), and utilisation (i.e., the quantity of healthcare services and procedures used). This model encompasses physical access, resource availability, cultural acceptability, financial affordability, and quality of care. More recently, Lévesque et al. (21) conceptualised access as the ability to identify healthcare needs, to seek services, to reach resources, to obtain or use services and to be offered services appropriate to needs. These, along with the various other conceptualisations of healthcare access (22-24), since the 1980s evince the complexity of the construct but also indicate the importance of examining the healthcare experiences of different groups in society.

In order to improve access to healthcare or the ‘fit’ between patients and healthcare services, a growing body of literature has emphasised the need to provide ‘reasonably adjusted’ health services and care to autistic individuals (25-27). In the UK, the Autism Act (2009) and the Adult Autism Strategy (2010) set forth guidelines for meeting the needs of autistic adults by improving the provision of services, including healthcare (28). However, the development and implementation of ‘reasonable adjustments’ and other interventions to improve healthcare services, requires an investigation and understanding of the specific challenges and barriers experienced by autistic individuals in healthcare settings (7). In a recent systematic review of self-reported barriers and facilitators to physical healthcare access for autistic adults, the authors recommend that further research is required which represents the perspectives of clinicians and caregivers/relatives as well as autistic adults in order to provider a fuller picture and that a robust framework of barriers should be developed (29). Accordingly, the aims of the current systematic review were to: 1) synthesise the extant research on barriers to physical healthcare as perceived by autistic individuals, their caregivers, and healthcare providers; and 2) use these data to develop a comprehensive taxonomy of the barriers to physical healthcare for autistic individuals. Taxonomies are tools that are commonly used in health services research to provide clear information on a phenomenon and inform appropriate measurement and/or assessment of quality and efficacy (30-33).

Methods

This review was conducted in accordance with the PRISMA guidelines (34), and the review protocol was registered on PROSPERO (the international prospective register of systematic reviews; registration number: CRD42018102372).

Inclusion & exclusion criteria

In order to meet the criteria for inclusion in this systematic review, it was required that: a) studies were focused on identifying the barriers to accessing appropriate physical healthcare for autistic individuals (for the purpose of this paper, physical healthcare was defined as any medical care related to the physical wellbeing of the participants); b) study participants were autistic individuals of any age, the parents/primary caregivers of autistic children or adults, or healthcare professionals who had experience of providing care to autistic individuals; c) studies were published in peer reviewed journals, and; d) studies were published in English. Mixed methods studies, quantitative studies and qualitative studies were included.

Studies were excluded if: a) they did not identify barriers to healthcare access for autistic individuals; b) they were focused on identifying barriers to healthcare access for persons with neurological conditions/disabilities other than autism, or if autism diagnosis was unclear; c) they did not report results for autism groups separately from other participant groups (e.g. intellectual disability, physical disabilities); d) they focused solely on dentistry or mental health services; and e) barriers to physical healthcare services could not be distinguished from barriers associated with other types of healthcare.

Search strategy

Studies were identified through systematic searches of five electronic databases: Medline (OVID), PsycINFO (OVID), CINAHL (EBSCOhost), Scopus (Elsevier), and the Psychology and Behavioral Sciences Collection (EBSCOhost). Initial searches were conducted in January 2018 and updated in July 2018. Searches used three distinct sets of search terms: 1) terms pertaining to Autism Spectrum Disorder (e.g., autism, autistic); 2) terms relating to primary and secondary healthcare services (e.g., physicians); and 3) terms relating to barriers to accessing appropriate and effective healthcare (e.g., barriers, disparities). Relevant medical subject headings (MeSH Terms) were used in Medline, CINAHL and PsycINFO in addition to free text key terms. The Medline search strategy is included as (Appendix 1.1). This search was adapted, where necessary, for the other databases. No restrictions were applied within searches regarding country or year of publication. Further, the reference lists of all studies identified as suitable for inclusion were also screened in order to identify additional, potentially relevant, studies.

Study selection

The search returns within each database were exported to an Excel file for screening. Within this file, one researcher (CW) screened all titles and abstracts against the inclusion and exclusion criteria. The full texts of all studies which appeared relevant were then examined by CW and a decision regarding inclusion was made. For any articles in which a decision about inclusion or exclusion was unclear, the research team (CW, SL, EOD & POC) discussed the article until consensus was achieved.

Data extraction

Data were extracted from each study independently by two reviewers (CW & EOD) on the following variables: study design; participant characteristics (i.e., *n*, age, type of participant – i.e., autistic individual/caregiver/HCP); healthcare setting; sampling methodology; country of publication; barriers identified. In order to assess inter-rater agreement, the individual barriers in each study that both reviewers had independently identified were examined, and the degree of agreement between the data extraction of the two reviewers was calculated (number of agreements/number of opportunities for agreement x 100). An initial agreement rating of 81.8% (range 42.37%-100%) was obtained. Any disagreements were resolved through discussion until consensus was achieved.

Quality assessment

The critical appraisal was conducted by two raters (CW & EOD) working together using the Quality Assessment Tool for Studies with Diverse Designs (QATSDD; 35). Two raters working together facilitates both verification and confirmability of the data by encouraging on-going dialogue between the raters (36). The QATSDD comprises of 16 items, each of which is scored on a scale ranging from 0 (not at all) to 3 (complete), with higher scores indicative of better methodological quality. A total QATSDD score was calculated for each study within this review and could range from 0 to 42 (quantitative and qualitative studies) or 48 (mixed methods studies). The QATSDD assesses a range of factors, including items of reporting adequacy (e.g., fit between research question and method of analysis), internal validity (e.g., statistical assessment of reliability and validity of measurement tools), and external validity (e.g., representative target group of a reasonable size). The QATSDD offers a useful means of comparing methodological quality among studies using different designs and has been used widely in systematic reviews focused on health services research (37-39). Any discrepancies in scoring between the raters were resolved through discussion and additional review of the paper. The quality appraisal was not used to inform the synthesis of results, however some papers contributed more to the review findings due to richness of data provided.

Thematic analysis

The barriers reported within the individual included studies were collated using a deductive thematic analysis approach. This is a method of identifying, reporting and analysing patterns (themes) within a dataset (40). This thematic analysis approach was guided by the process described by Braun and Clarke (40):

1. Familiarisation with the data: two reviewers (CW & EOD) independently read all of the included papers several times, making broad notes about the barriers reported within each.
2. Generating initial codes: each individual barrier identified in the included studies (e.g., waiting times, loud noises, lack of healthcare provider (HCP) knowledge) was treated as an individual code. For qualitative data, barriers were derived from direct quotes from study participants or author report based on interviews/focus groups. For

quantitative data, all barriers in each survey, that were endorsed by participants, were coded as barriers.

3. Searching for themes: Raymaker and Colleagues (41) previously produced a questionnaire in which they organised potential barriers to healthcare for autistic individuals under nine categories. These were: Emotional, Executive Function, Healthcare Navigation, Provider Attitudes, Patient-Provider Communication, Sensory, Socio-Economic, Support, Waiting, and Examination Rooms. For the purpose of this review, these categories were treated as initial, pre-determined, themes. Raymaker and colleagues' (41) categories were developed through a community-based participatory research approach whereby academics and members of the autism community served as equal partners throughout the research process. Therefore, these categories were seen as an insightful guide to the current analysis. Four reviewers (CW, SL, EOD & POC) examined the individual barriers reported in the included studies against these pre-determined themes. Themes were modified, or new themes were developed whenever individual barrier codes identified within the papers did not fit well into the pre-specified themes. The original texts from which the individual barrier codes had been retrieved were referred to throughout the process to ensure the correct context of the code was retained.
4. Reviewing the themes. The research team met a second time to review the codes and themes to ensure a good fit for the data.
5. Defining and naming themes: the research team then organised the themes into an overall taxonomy of themes and subthemes. Each theme was defined, and final overall theme names were decided.

A qualitative narrative synthesis was employed for reporting the results. Narrative synthesis is used to summarise and describe findings primarily through words and text as opposed to statistical data (42). A qualitative synthesis was chosen for this review because qualitative research is well suited to understanding the complex phenomena that exist in healthcare and for revealing links between them (43).

Results

Search results

Figure 1 presents the PRISMA flow diagram for this review and depicts the number of studies considered by the researchers at each stage of the search and review process. Database searches returned a total of 12,957 potentially relevant articles and ultimately a total of 31 studies were found to meet the inclusion criteria. The included studies were published between the years 2003 to 2018, with the majority published since 2015 (*n*=21; 67.74%). A detailed study-by-study summary of included studies is presented within Appendix 1.2.

Study design

Of the included studies, 14 were solely quantitative in nature, 15 employed qualitative research methods only and three studies reported the use of a mixed or multi methods approach. Specific research methodologies utilised included surveys/questionnaires (*n*=16; 51.61%), interviews (*n*=13; 41.93%), focus groups (*n*=3; 9.67%), pre-healthcare visit telephone assessments (*n*=1; 3.23%), collaborative meetings and guidelines review with autistic individuals and professionals working in the area of autism (*n*=1; 3.23%), and a clinical vignette describing an autistic child (*n*=1; 3.23%; see Appendix 1.2).

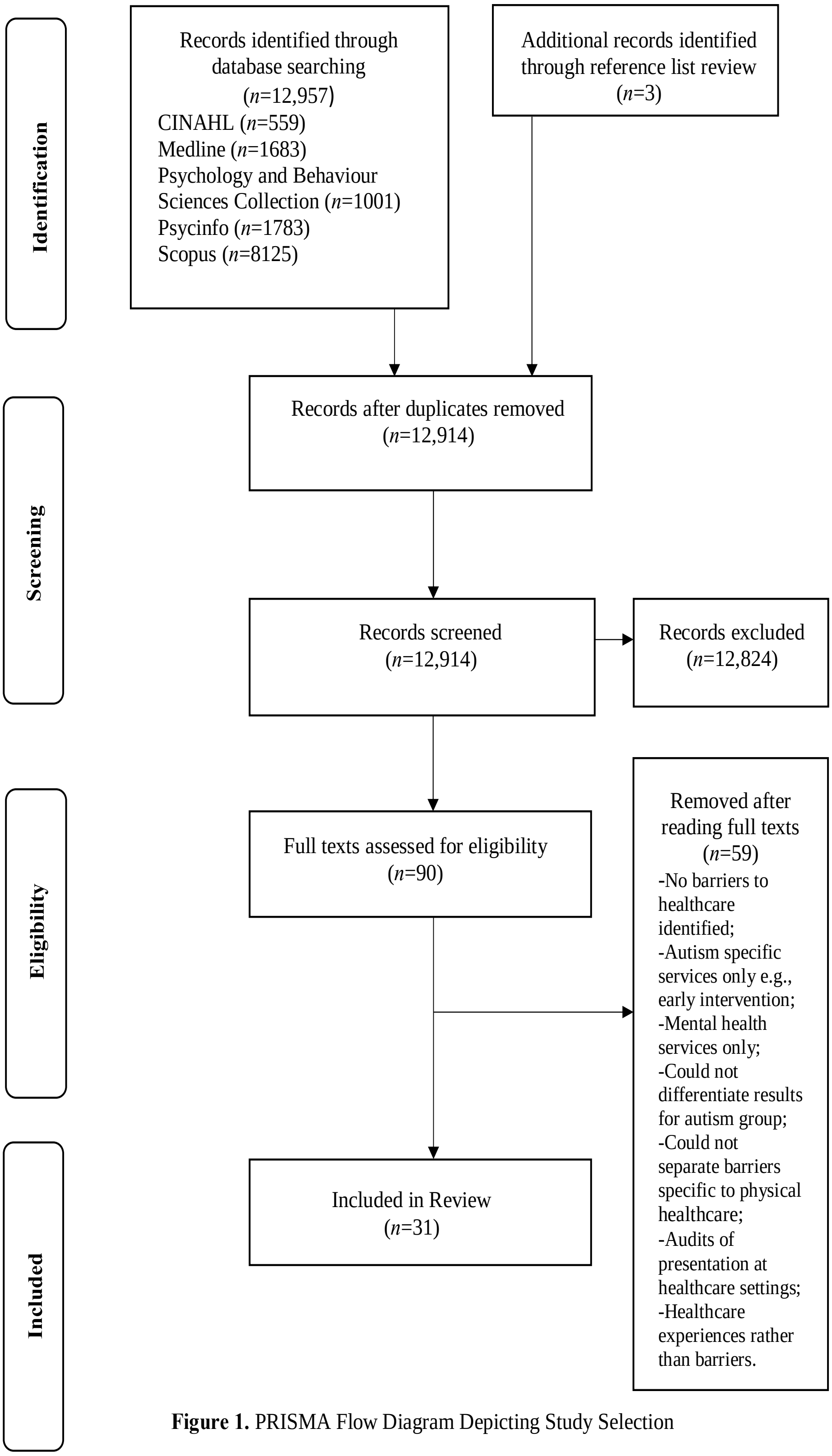

Figure 1. PRISMA Flow Diagram Depicting Study Selection

Study participants

Autistic individuals participated in seven of the 31 studies reviewed (22.58% of studies; *M*=50 autistic individuals per study, *SD*=79.22, range: *n*=6-209; Dern and Sappok (44) did not provide this information on participant characteristics). Of these, autistic adults were participants in five studies (age 18+; 16.13% of all studies; Dern & Sappok (44) was not included in the calculations as they did not provide this information), and autistic children/youth (younger than 18 years) participated in two studies (6.45% of all studies). Caregivers participated in 18 (58.06%) of the 31 studies (*M*=29.79 caregivers per study, *SD*=46.03; range: *n*=6-183). Caregiver characteristics were not specified in four studies (45-48), but of the studies that did, most caregivers were the parents of an autistic individual (*n*=13 studies; 41.93% of studies; *M*=30.85 parents per study, *SD*=47.74, range: *n*=6-183), and the majority were mothers (*n*=11 studies; 35.48% of studies; *M*=26.18 mothers per study, *SD*=46.11, range: *n*=5-164). HCPs participated in 14 studies (45.16%; *M*=199.81 HCPs per study, *SD*=316.66, range: *n*=10-1163). The participating HCPs were most commonly physicians (*n*=14, 45.16% of studies; *M*=192.2 physicians per study, *SD*=322.86, Range: *n*=4-1163). A combination of participants from two or more of the three groups (i.e., autistic individuals, caregivers, HCPs) participated in nine studies (29.03% of all studies; see Appendix 1.2).

Sampling methods

As can be seen in Appendix 1.2, convenience sampling, such as approaching participants who were attending a clinic to participate in research (e.g., 49), was the most common type of sampling method used across the included studies (*n*=20; 65.5%). Other types of sampling included snowballing methods (*n*=4; 12.9%), such as through word of mouth (e.g., 50), and purposive sampling (*n*=4; 12.9%), such a purposively targeting members of a specific professional group (e.g., 51). A number of studies (*n*=5; 16.13%) drew their sample from a larger study, for example, Lake et al. (52) invited caregiver participants who were taking part in a larger ongoing study examining health care utilisation patterns among autistic adolescents and adults.

Healthcare Settings

There was some overlap in terms of healthcare settings as some studies looked at more than one setting, but primary care settings were the focus of the majority of studies (*n*=15; 48.39%). Secondary care was the focus of four studies (12.9%) and the emergency department was considered in three studies (9.68%). Appendix 1.2 provides more information on the variety of healthcare settings covered by the included papers.

Countries of origin

The majority of the included studies were conducted in the USA (*n*=22; 70.97%). The other studies were conducted in Canada (*n*=4; 12.9%), Sweden (*n*=1; 3.23%), the UK (*n*=1; 3.23%), Germany (*n*=1; 3.23%), Australia (*n*=1; 3.23%) and Nigeria (*n*=1; 3.23%; See Appendix 1.2).

Quality assessment

The overall mean QATSDD score across all studies was 21.7 (*SD*= 4.43, range: 6-28) out of a maximum possible score of 42 for quantitative and qualitative studies, and 48 for mixed methods studies. Quantitative studies alone scored an average of 21 (*SD*=2.8; *n*=14; range:16-26) out of a possible 42. Qualitative studies (*n*=15) scored an average of 22 (*SD*=6; range: 6-28) out of a possible 42. All three mixed methods studies scored 26 out of a possible 48. Studies scored most poorly on items related to referring to a theoretical framework, adequately reporting sample size considerations or providing detailed recruitment data and demonstrating clear user involvement in study design. However, the included studies performed well on items relating to providing clear aims and objectives, presenting a clear description of the research setting and ensuring a good fit between the research question and method of analysis.

Synthesis of identified barriers

A total of 320 individual barriers were reported across the included studies (*M*=10.32 barriers reported per study, *SD*=6.76, range 4-37), with overlap evident, and were suitable for analysis and categorisation. The categories of barriers to healthcare described by Raymaker and colleagues (41) were used as an initial framework. However, there were a substantial number of barriers reported within the included studies that could not be classified under the categories within this framework, such as parents feeling dismissed by staff (53), or a lack of consistency or collaboration between services/HCPs (54). Therefore, changes were made to the existing themes or new themes were created in order to revise this initial framework into a new taxonomy.

Themes

The thematic analysis ultimately resulted in the emergence of a taxonomy of four themes with each consisting of between two and eight subthemes. The taxonomy, which collectively presents the themes and subthemes of barriers reported by autistic individuals, their caregivers, and HCPs, is presented in Figure 2. The themes and subthemes are also presented with illustrative examples in Table 1.

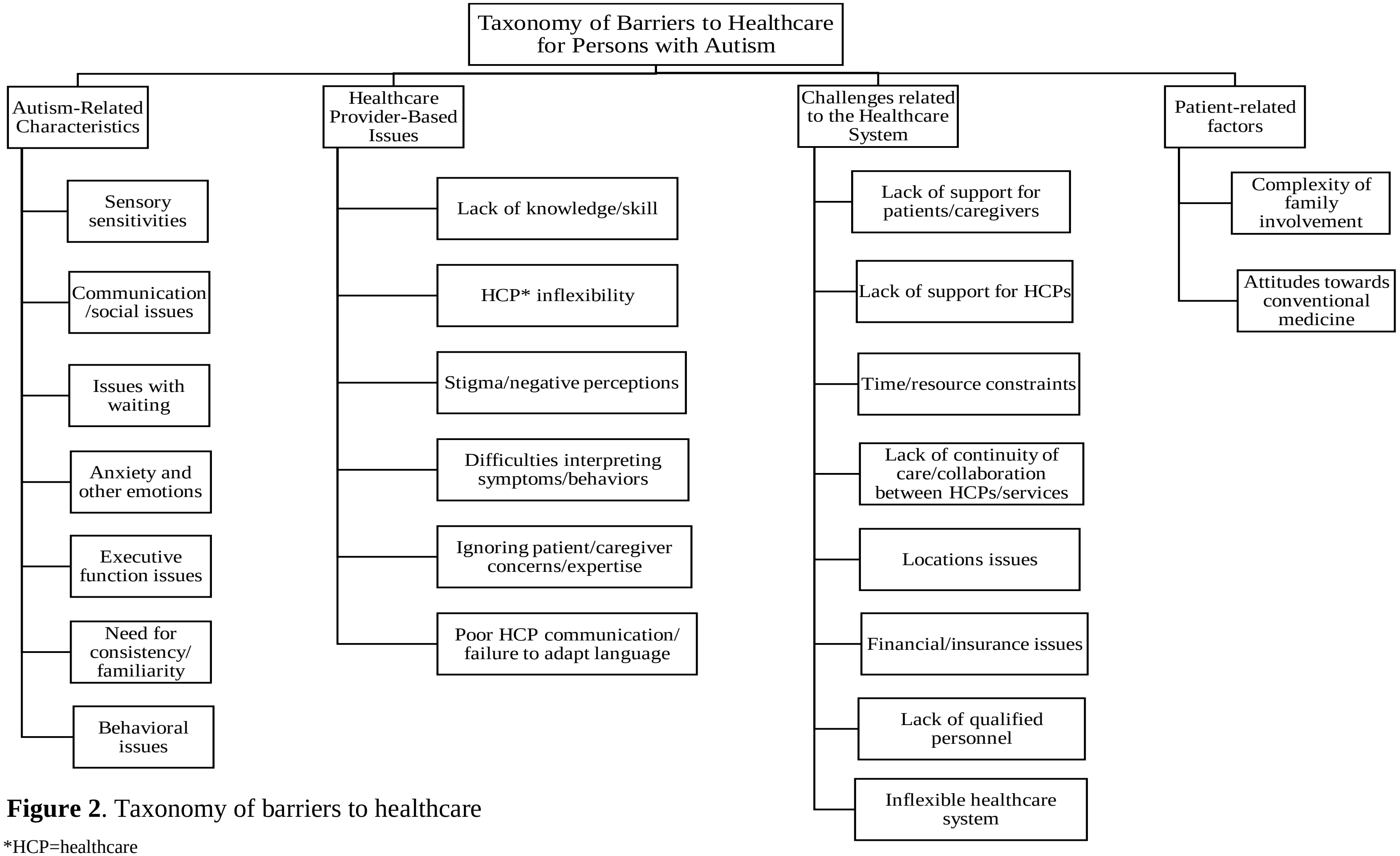

Figure 2. Taxonomy of barriers to healthcare

*HCP=healthcare

Table 1. Themes and subthemes with illustrative examples

Theme **(*n* studies)**	**Subtheme** **(*n* studies)**	**Illustrative Examples**
Autism-related characteristics (*n*=17)	Sensory issues (*n*=8)	- '*Well since I'm really sensitive to noise, it was really hard*'– Autistic individual; qualitative study (55). - '*He doesn't like loud noises…if most things are quiet, he can handle it, but if there's so much noise going on at once, it bothers him a lot*' – Caregiver; qualitative study (54). - '*There's so many machines, high-pitched squeals, not good lighting, people talking, lots of people milling about, walking by, it's just everything that you would not want potentially as an individual with autism would be there*' HCP; qualitative study (56).
	Communication and social issues (*n*=14)	- "*'It is hard for me because I don't have the words that normal people have to communicate with. I don't always know how to respond properly to questions from healthcare providers*' – Autistic individual; qualitative study (57). - *"…if we can minimise interactions, he doesn't get upset before the surgery*" – Caregiver; qualitative study (54). - '*It is very difficult because it is more like doing medicine for an infant because he can't tell me the severity of his feelings…and definitely he can't tell me any history…*'- HCP; mixed methods study.[58]
	Issues with Waiting (*n*=14)	- Specific barriers included the waiting room and waiting in general– Autistic individual (author report); mixed methods study (48). - '*That's the only thing I would change, just the length of wait time before surgery because she gets so anxious'* – Caregiver qualitative study (49). - '*With the waiting beforehand they (autistic individuals) start escalating their behaviours and they sometimes get agitated and combative…that's when it starts to get a bit tense in the environment'* - HCP; qualitative study (59).

	Anxiety & other emotions (*n*=6)	- The anticipation of social interaction with the medical team and overstimulation created increased stress for autistic individuals which in turn inhibited their concentration and ability to interact with the HCP – Autistic individual (author report); qualitative study (48). - Child fears about the examination/doctor were endorsed as a barrier to providing care by 67%/47% of HCPs (*n*=23); quantitative study (60). - ‘*He is not needle-friendly at all – he gets wicked anxiety*’ – Caregiver; qualitative study (53).
	Executive function issues (*n*=3)	- ‘*With my autism it is very difficult for me to understand and follow all the different appointments and procedures I have to schedule and how to do it*’ - Autistic individual; qualitative study (57). - Not admitting to hospital because of rigid thinking (e.g., not being able to water plants at home and not seeing an alternative) – Autistic individuals/professionals working with autistic individuals (author report); qualitative study (44).
	Need for consistency/ familiarity (*n*=6)	- A need for consistency was noted as a barrier to care – Autistic individual (author report); qualitative study (57). - *‘His autism is most noticeable when he has to face change or transitions*’ – Caregiver; qualitative study (55). - ED was described as a place that optimally functioned when issues were dealt with quickly and by following procedures that were discordant with the child’s preference for their own routines and familiar materials – HCPs (author report); qualitative study (56)
	Behavioural issues (*n*=7)	- Child behaviour during encounter being a challenge was endorsed as a barrier by 78% (*n*=38) of HCPs and 24% (*n*=11) of caregivers; quantitative study (60). - ‘*My biggest fear is the kid that’s more aggressive*’ – HCP; qualitative study (61).
Provider-Based Issues (*n*=24)	Lack of provider knowledge/ Skill (*n*=17)	- ‘*I have gotten the distinct impression that all of the physicians I have seen have no clue what autism means or entails or how that should change how they should treat me’* – Autistic individual; qualitative study (57). - ‘*I felt I had to educate the doctors regarding my son’s autism*’ – Caregivers; qualitative study (54).

		- *‘There was virtually no education or supervised experience for me (around working with autistic individuals) during my residency, and although that’s a little bit better today, it is still far from being where it needs to be*’ – HCP; qualitative study (52).
	HCP inflexibility/ unwilling to make accommodations (*n*=12)	- ‘*I prefer and find it easier to communicate in text…but with every doctor I speak to, they wave away the note card and look at me to ask the same question I have just answered…I wish healthcare providers would read the notes I write for them*’ – Autistic individual; qualitative study (57). - ‘*I have to ask them before they go in the room “can you please remove your coat?” if they don’t and they’re like “no, he’ll be fine”, it’s hold down, it’s scream “no, no mommy, no…doctors are not receptive to the request”* – Caregiver; qualitative study (62).
	Stigma/ negative perceptions (*n*=5)	- *‘I am very careful about disclosing my autism diagnosis to my healthcare provider because I fear it’s gonna affect my healthcare*’ – Autistic individual qualitative study (57). - *I’ve taken my son to a doctor who just really didn’t know how to deal with him, so he told us not to come back’* – Caregiver; qualitative study (54).
	Difficulties interpreting symptoms/ behaviours (*n*=2)	- Staff don’t believe me when I tell them that new symptoms are not related to an existing condition/disability - statement endorsed by 19% of autistic adults; quantitative study (41). - Interpreting symptoms was complicated by autism-related behaviours – HCPs/caregivers (author report); qualitative study (63).
	Ignoring patient/carer concerns/ expertise (*n*=9)	- ‘*The triage person kept speaking to the person who brought me in rather than me. The lady could have spoken to me*’ – Autistic individual; qualitative study (57). - A child reported that doctors in the emergency department ignored him despite the child asking many questions due to being nervous – Autistic individual (author report); qualitative study (53). - Frustration or anger were felt when caregivers felt personal expertise of their own child was not respected or their concerns were ignored – caregivers (author report); mixed methods study (50).
	Poor HCP communication/	- ‘*I could only know a bit of what they were saying. I don’t know much of the grown-up words in English, I only know the basic words of English*’ – Autistic individual; qualitative study (53). - ‘*They spoke to him as if he was hard of hearing*’ – Caregiver; qualitative study (64).

	failure to adapt language (*n*=6)	
Healthcare System-Based Issues (*n*=24)	Lack of supports available for patients/ caregivers (*n*=9)	- *‘I wish they understood how easy it is to get confused…I need someone to hold my hand…there is definitely nobody willing to do that*’ – Autistic individual; qualitative study (57). - *‘I experienced that I had no support anywhere’* – Caregiver; qualitative study (64). - *‘Support for families going through this process is lacking, with delays, buck passing and frustration all around*’ – HCP quantitative study – statement from opened-ended survey item (51).
	Lack of supports available for HCPs (*n*=9)	- ‘*The resources for supporting GPs are poor’* – HCPs; quantitative study – statement from opened-ended survey item (51). - A lack of practice guidelines was endorsed as a barrier to caring for autistic children by 67% of participating HCPs; quantitative study (65).
	Time/resource constraints (*n*=11)	- ‘*Everything is so time limited, you get going and you feel sort of secure…and school ends and everything falls apart again’* – Caregiver; qualitative study (52). - ‘*If it’s really crazy busy…and I just can’t, or I don’t have the time or effort to devote that I might have on a quiet day, then sometimes it’s hard*” – HCP; qualitative study (56).
	Lack of continuity of care/ collaboration between HCPs (*n*=19)	- ‘*There are so many people on her caseload, and they don’t communicate with each other*’ – Caregiver; qualitative study (59). - ‘*With kids who have a number of chronic issues… (the staff) have to do a transition process…I don’t think (that) kids with autism…are being followed in the same way (with) transition (to adult care)*’ – HCP; qualitative study (61).
	Location issues (*n*=2)	- No provider available in the area was endorsed as a barrier to routine preventative care for 3.7% of Caregivers (*n* not specified; author report); quantitative (46). - Living in rural area/doctor’s office being too far away was endorsed by 12% (*n*=25) of autistic individuals; quantitative study (41).

	Financial and insurance issues (*n*=11)	- Concern about cost of care keeps me from getting primary care – statement endorsed by 30% (*n*=62) autistic individuals; quantitative study (41). - The cost of routine preventative care was endorsed as a barrier by 28.4% of caregivers of 2,088 autistic children; quantitative study (46). - ‘*Because they (autistic individuals) take more time and the more time you spend with a patient, the less money you’re making’* – HCP; qualitative study (66).
	A lack of appropriately qualified personnel (*n*=5)	- The most commonly endorsed provider-based access problem was finding skilled and experienced specialty doctors - 18.4% of Caregiver participants (*n* not specified; author report); quantitative study (47). - A dearth of speech and behavioural therapists was endorsed as a barrier by 100% (*n*=19) of participating HCPs; quantitative study (67). - A lack of access to autism specialists was endorsed as a barrier by 64% (*n*=9) of participating HCPs; quantitative study (68).
	Inflexible healthcare system (*n*=5)	- ‘*It really doesn’t take a whole lot to modify things so that you can meet the needs for most of the people on the spectrum. Right now, those offices are set up for the physicians, they are not set up for the patients*’ – Autistic individual; qualitative study (57). - ‘*The staff did what they could, but different routines are required for a girl like my daughter’*– Caregiver; qualitative study (64). - ‘*We give (autistic patients) the antithesis of what they need…a hospital is a very inflexible place…it doesn’t adapt itself to the people who are in it*’ – HCP; qualitative study (55).
Patient-related factors (*n*=9)	Complexity of family involvement (*n*=5)	- ‘*Well, they (autistic patients) are much more likely to have, almost 100% more likely to have someone with the patient…so right away you have two patients…so that’s very different and that’s something that a lot of internists aren’t used to’* – HCP; qualitative (66). - Difficulties relating to guardianship was endorsed as a barrier by 12.1% of 183 caregivers; quantitative study (69).

	Attitudes towards conventional medicine (*n*=5)	- The family being sceptical of vaccines was endorsed as a barrier to providing care to autistic individuals by 73% (*n*=92) of HCPs; quantitative study (65). - *'Some doctors can be a little more resistant to discussing this (alternative treatments for autism)'* – Caregiver; qualitative study (62). - Parent beliefs about healthcare was endorsed as a barrier by 27% (*n*=13) of HCP; quantitative study (60).

GP=general practitioner; HCP=healthcare provider

Theme 1: Challenges associated with autism-related characteristics

All participant groups (i.e., autistic individuals, caregivers, HCPs) reported or endorsed barriers to healthcare that were related to characteristics of autism, such as sensory sensitivities or communication and social difficulties (*n*=17 studies; 54.84%; for illustrative examples, see Table 1). For example, bright lights or loud noises were noted as sensory triggers that caused discomfort for autistic individuals in healthcare settings (e.g., 49,53,54). In addition, HCPs sometimes found it difficult to communicate with patients who may have limited verbal abilities (e.g., statement endorsed by 55% of respondents, *n*=27; 60), while autistic individuals reported that they sometimes had difficulty articulating their symptoms to HCPs (e.g., 55). Difficulties handling waiting rooms and lengthy waiting times were another subtheme that all three participant groups noted or endorsed (*n*=14; 45.16% studies; see Table 1).

Theme 2: Healthcare provider-related issues

HCP-related issues were reported in 24 studies (77.42%; for illustrative examples, see Table 1). This theme captured barriers to accessing healthcare that were perceived to be related to HCPs. A common barrier reported by both caregivers and HCPs was a lack of HCP knowledge and training regarding autism (e.g., 46, 58). As a result, HCPs often reported that they had low self-efficacy in managing the medical care of autistic individuals (e.g., statement endorsed by 43% of participants, *n*=6; 68). Caregivers and autistic individuals also highlighted a lack of flexibility or an unwillingness by the HCP to make accommodations for the patient (e.g., 57, 62). Perceived stigma towards autism among HCPs was another subtheme reported by caregivers and autistic individuals. Caregivers sometimes reported feeling judged by other parents and staff because of their child's behaviour (50). Further, some autistic individuals noted that they were sometimes reluctant to disclose their diagnosis for fear it would negatively impact their care (e.g., statement endorsed by 75% of participants, *n*=15; 70). Table 1 presents the subthemes for this theme with illustrative examples.

Theme 3: Challenges related to the healthcare system

Barriers related to the healthcare system were also experienced by autistic individuals, caregivers, and HCPs (*n*=24 studies; 77.42%; for illustrative examples, see Table 1). For caregivers and autistic individuals, these were mainly related to a lack of available supports both for accessing and navigating the healthcare system (e.g., 52,57). HCPs also felt that there was a lack of guidelines available to HCPs for caring for autistic individuals (statement endorsed by 48% of participants; *n*=259; 71). A general lack of resources within the healthcare system was also a barrier for HCPs; for example, administrative staff shortages and a general lack of time to manage the care of autistic individuals effectively were reported (e.g. statements endorsed by 36.7% [*n*=335] and 38.6% [*n*=319] respectivley; 72). Parents and HCPs also reported difficulties in relation to continuity of care or collaboration between staff and services (54, 63). Also within this theme were issues relating to the location of healthcare services and the financing of, and payment for, services. Statements regarding location issues were endorsed by autistic individuals and caregivers and were mainly in relation to living too far away from the healthcare centre or having difficulties accessing transport to attend the healthcare service or centre (41,46). Financial/insurance issues (*n*=11 studies; 35.48%) were reported or endorsed by all three groups but varied in nature by group. For HCPs, financial issues were related to a lack of reimbursement, often associated with the extra time required for autistic individuals during consultations (statement endorsed by 14% of participants, *n*=2; 68). For caregivers and autistic individuals, on the other hand, difficulties with insurance coverage and the cost of healthcare were the main concerns (e.g., 41,54).

Theme 4: Patient-related factors

Subthemes in theme four were mainly reported by HCPs (*n*=9 studies; 29%; see Table 2 for illustrative examples). This theme encompassed challenges with healthcare access which are related to the individual and included complexities of family involvement in care, and attitudes regarding conventional medicine among autistic individuals and caregivers. HCPs felt that having caregivers present with the patient changed the dynamic and length of the consultation as they were now effectively interacting with two patients and as a result, explanations could take longer (58). The family's lack of acceptance of autism was also noted as creating issues (statement endorsed by 79% of participants, *n*=15; 67).

Care could be further complicated by attitudes towards healthcare by autistic individuals and caregivers. For example, HCPs in four studies (12.9%) endorsed statements regarding families' use of complementary alternative medicine (CAM) and being sceptical of vaccines as barriers to providing care to autistic individuals (60,65,71,81). Caregivers, on the other hand, expressed a desire for HCPs to be more open to discussing alternative approaches to treating autism (62).

Differences across groups

Figure 3 shows the distribution of the themes and subthemes reported by each participant group according to the number of studies in which they were identified. For autistic individuals, the most common subthemes were related to communication and social issues, and HCPs' failure to adapt language, while the most common subtheme reported by HCPs was a lack of continuity of health care/collaboration between services/HCPs. This was also the most common subtheme according to caregivers, followed closely by issues with waiting.

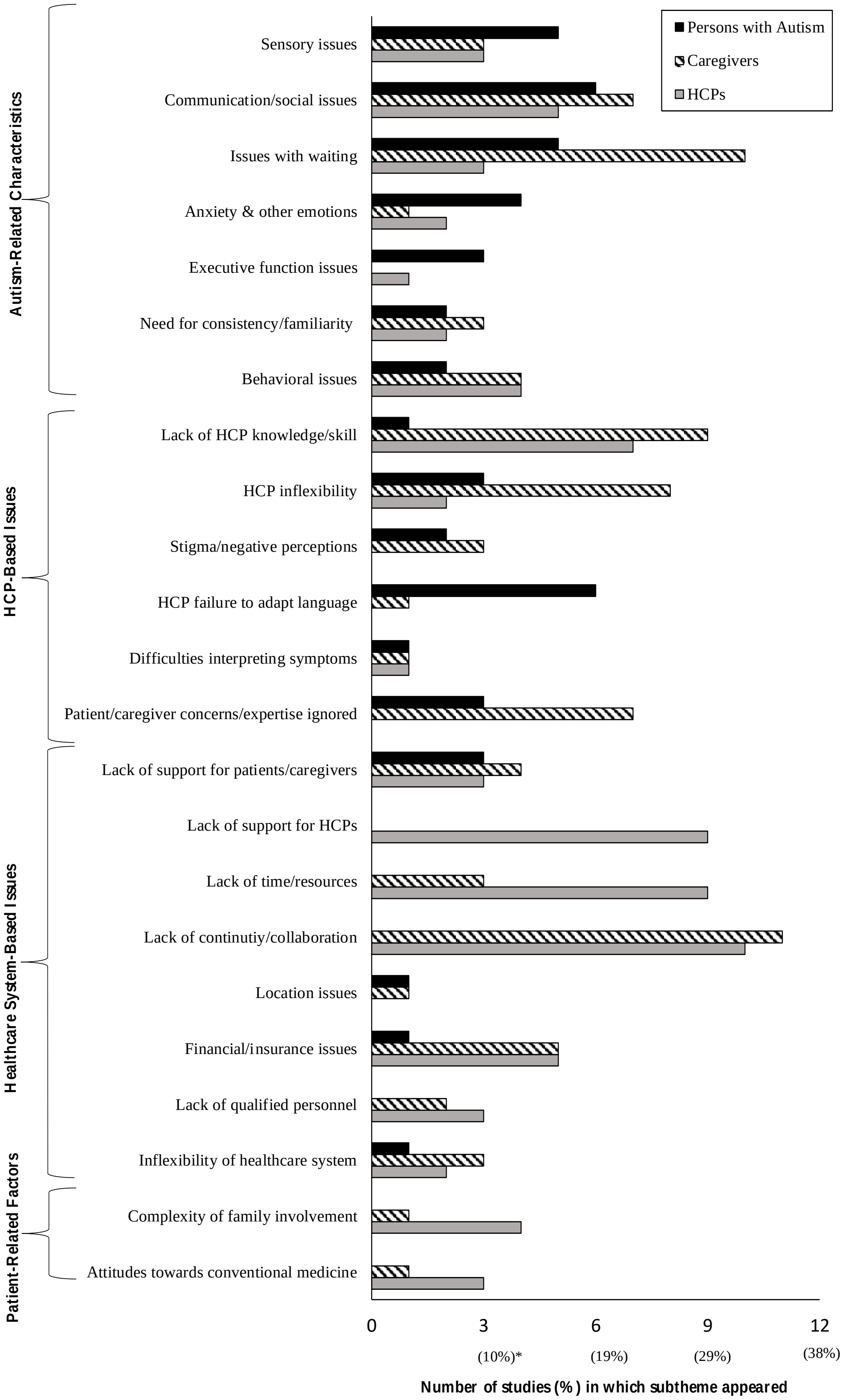

Figure 3. Frequency of themes and subthemes reported by autistic individuals, caregivers, and HCPs
HCP=healthcare provider

Discussion

Summary

This systematic review has allowed for the development of a comprehensive taxonomy that can inform service evaluation and improvement of healthcare for autistic individuals. It has also elucidated differences between autistic individuals, their caregivers, and HCPs with regards to perceived barriers and deficits in healthcare provision. The barriers reported by autistic individuals, caregivers and HCPs in the various studies were organised into four overarching themes: Barriers Associated with Autism-Related Characteristics; HCP-related; Healthcare System-related Issues; and Other Patient-Related Factors.

The current findings indicate that barriers to healthcare for autistic individuals occur at the patient, HCP, and system levels. This echoes findings from healthcare accessibility research within other vulnerable populations. In examining barriers to reasonably adjusted care, Tuffrey-Wijne et al. (84), for example, found that persons with intellectual disabilities, their caregivers and healthcare staff also reported barriers at the patient (e.g., lack of ability to ask for adjustments to be made), staff (e.g., lack of staff knowledge about reasonable adjustments), organisational (e.g., lack of senior management support) and cross-organisational levels (e.g., patients with intellectual disabilities not being identified and flagged).

The Equality and Human Rights Commission (85) define three categories of reasonable adjustments that can be applied to healthcare settings: 1) adjustments to physical features; 2) auxiliary aids and services; and 3) adjustments to policies and procedures. Numerous studies have shown that implementing such adjustments can be beneficial to persons with developmental disabilities in healthcare settings. For example, hand-held health records, such as hospital passports, have been shown to facilitate discussion about health problems, and increase health-related knowledge and awareness of personal health issues among people with intellectual disabilities (86). Similarly, adapting the sensory environment has been shown to reduce anxiety, reported pain and discomfort for autistic children attending the dentist when compared to a standard dental environment (87). However, there is first a need to understand what specific reasonable adjustments are required for individual patients. Future research synthesising work on reasonable adjustments and linking these to specific categories of barriers is necessary to advance care.

The taxonomy developed herein offers clear and consistent language and terms that can be used to describe the barriers to healthcare for autistic individuals. Across the included studies, there was no standardised labelling of barriers and the precise nature of many barriers was unclear without in-depth examination. Additionally, varying language was often used to describe the same barrier (e.g., parents feeling dismissed by staff; parents' concerns ignored; parents' expertise not being respected; HCPs not listening to parents). Through a thematic analysis approach, the research team was able to collate the various barriers, and the terms used to describe them, into one clear taxonomy. Such taxonomies have been found to be useful in other fields of healthcare to identify areas in need of intervention and to aid in intervention development (88, 89). Therefore, this taxonomy may also spur improvement efforts relating to healthcare services for autistic individuals as HCPs have reported that not knowing what accommodations were needed by patients with developmental disabilities was an impediment to implementing accommodations in clinical practice (84). Finally, a wide variety of tools and methods were used in the included studies to assess perceived barriers. However, no one tool was sufficient to capture all of the barriers which have been identified in this review by autistic individuals, caregivers, and HCPs. It is therefore hoped that this taxonomy will also facilitate the development of more comprehensive tools for assessing perceived barriers within healthcare settings.

Addressing barriers associated with autism-related characteristics. Interventions targeting barriers under the theme of Autism-Related Characteristics should focus on making accommodations that would reduce frequently occurring barriers such as communication and social difficulties, difficulties with waiting and sensory sensitivities. Table 2 presents examples of approaches which may be helpful in addressing the three most commonly reported subthemes under this theme. The level of empirical evidence to support the suggested approaches in this table varies and future research is required to establish the efficacy and utility of these for addressing specific barriers to healthcare encountered by autistic individuals. Although some of the individual barriers within this theme are similar to what has been reported in the wider developmental disability literature (90,91), it is important to note that there are nuances in the experience of these barriers among autistic individuals. For example, Raymaker and colleagues (41) found that communication issues can be a concern for persons without a comorbid intellectual disability. This emphasises the need for clinicians to be aware of the need for adjustments to care for all autistic individuals, regardless of where on the spectrum they fall. Similarly, the sensory sensitivities experienced by autistic individuals can cause significant challenges in healthcare settings that persons with other disabilities may not experience (41). There may also be a complex interplay between barriers that should be considered by future research. For example, a number of studies in this review suggested that anxiety related to the healthcare visit, or sensory discomfort caused by the healthcare environment, may negatively impact the patient’s ability to communicate with the HCP (41,57,70). These findings echo those of Mason and colleagues (29) in their systematic review of barriers and facilitators to healthcare experienced by autistic adults.

Table 2. Examples of potential approaches to address commonly reported barriers to care for autistic individuals.

	Barrier	**Potential Solution**	**Example**
Barriers associated with autism-related characteristics	Sensory sensitivities	Toolkits which allow autistic individuals /caregivers to notify HCPs in advance of any sensory issues they may have	The Autism Healthcare Accommodations Tool (AHAT), part of the AASPIRE Healthcare Toolkit (73), offers autistic individuals /caregivers a list of potential adjustments/accommodations (e.g., low lighting, quiet space) and allows them to choose which ones would be most useful to them, thereby creating an individualised report. This can be sent to the HCP in advance of the visit (98).
	Communication difficulties	Use picture/visual schedules before and during medical encounters to aid communication and decrease anxiety	Chebuhar et al. (74) developed picture schedules for a variety of medical procedures conducted in hospital settings. The majority of staff and caregivers who participated in the pilot study felt that using the picture schedules decreased child anxiety and maladaptive behaviours during the procedures.
	Issues with waiting	Give specific appointment times which suit the patient (e.g., first of the day/during or after lunch)	Pratt et al. (75) describe an initiative to improve hospital admission for persons with learning disabilities and autism. Their strategies include: admitting children to hospital as late as possible, and discharging as early as possible, even if this means at irregular times, so that they are not waiting around and nil by mouth time is minimised. Feedback from families regarding these strategies was positive.
HCP-related barriers	Lack of provider knowledge/ skill	Development and delivery of training programs	Major et al. (76) conducted a feasibility study of ‘Autism Case Training: A Developmental Behavioral Pediatrics Curriculum’. The curriculum consists of seven case-based teaching modules on autism-related content including signs and symptoms, screening, and treatment. Post-tests showed significant increases in recognising early warning signs of autism, and self-assessed autism knowledge and proficiency among paediatric residents.

	Patient/caregiver concerns/ expertise being ignored	Improve competency in shared decision making (SDM)	Bieber et al. (77) describe a general SDM physician training program which could be delivered in two four-hour sessions. The program was positively received by physicians, and increased physicians’ confidence to engage in SDM.
	HCP Inflexibility/ unwilling to make accommodations	OSCEs/ Simulated patients	McIntosh et al. (78) reported on how a simulated autistic patient was incorporated into the nursing curriculum. Student feedback indicated improved communication abilities and greater recognition of the need to make accommodations such as simplifying the environment, removing unnecessary stimulation such as light/sound and expediting the patient’s discharge to alleviate stress.
Healthcare system-related barriers	Lack of continuity/ collaboration	Develop and implement a shared plan of care	Clark et al. (79) describe a pilot study of the Linked Program which encourages communication and collaboration between caregivers of autistic children and HCPs and may facilitate smoother transitions through the surgical care process.
	Lack of support for HCPs	Develop guidelines	Crowe and Salt (80) summarise the different ways HCPs can support and care for young autistic people in the National Institute for Healthcare and Care Excellence (NICE) guidelines on ‘Autism: the management and support of children and young people on the autism spectrum’.
	Lack of resources	Training to improve HCP self-efficacy in treating autistic patients	Carbone et al. (81) conducted a learning collaborative training program with three cohorts of HCPs over 3-6 months (varied by cohort). This involved either a full- or half- day workshop or webinar, in which evidenced-based recommendations for autism surveillance and screening were discussed. Teams participated in monthly conference calls on a variety of autism-related topics. Post intervention surveys showed a significant improvement in self-efficacy and

			in a variety of resource related areas such as available time and care coordination services.
Other patient-related barriers	Complexity of family involvement	Longer appointment times would allow HCPs to provide full explanations and address any caregiver concerns or questions.	The National Autistic Society have prepared guidelines for health professionals which are available online (82). One such recommendation is to book double appointments for autistic patients.
	Attitudes towards conventional medicine	Increase HCP knowledge of evidence and non-evidence-based interventions so that they can provide appropriate information to families and patients.	Bordini et al. (83) describe a training program for primary care physicians which consisted of five three-hour sessions. Each session comprised two hours of lectures and one hour of case discussion. The themes of the sessions included autism epidemiology, symptoms and early signs of autism, diagnostic criteria, and evidenced-based treatments for autistic individuals. Knowledge of autism, including evidence-based treatments, increased significantly after the training program.

HCP=healthcare provider; OSCE=objective structured clinical exams;

Addressing healthcare provider-related issues

The most common subthemes relating to the HCP were a lack of HCP knowledge and skill regarding autism, patient/caregiver concerns or expertise being ignored, and HCP inflexibility or unwillingness to make accommodations. The lack of knowledge and training about autism as a barrier is unsurprising as there is substantial extant research suggesting that knowledge of autism among HCPs is poor (e.g., 92-95). However, research is beginning to show how this can be addressed in a variety of ways, including training programs and employing simulated patients who are on the autism spectrum. Table 2 provides examples of interventions which may be useful in addressing some of these barriers, though further evaluation of these interventions is required.

Caregivers/autistic individuals feeling that their concerns and expertise were being ignored by HCPs, and that they were, consequently, not being treated as partners in care is also consistent with the extant autism literature (96). Engaging in shared decision making has been shown to be significantly associated with improved satisfaction with the child’s overall primary healthcare for parents of autistic children and also with improved guidance regarding controversial treatments (97). However, when compared to children with Down’s Syndrome or Cerebral Palsy, autistic children were less likely to receive a shared decision-making approach to care (98). More collaborative healthcare provision and planning, which involves the caregiver and/or patient, could also potentially reduce the occurrence of diagnostic overshadowing (i.e., the misinterpretation of symptoms as being related to an existing condition rather than being signs of an undiagnosed medical issue; 99). Diagnostic overshadowing is commonly reported by autistic individuals and individuals with other developmental disabilities (e.g., 84, 100) and may be the result of HCPs having difficulties in interpreting symptoms in their autistic patients (63). Caregivers are likely better equipped to provide important information regarding new/unusual behaviours that may signal a physical health issue, particularly when it is difficult for the patient to articulate their pain/symptoms (100).

Addressing healthcare system issues

At the systemic level of the health services, subthemes identified included a lack of continuity of care, the rigidity of the healthcare system and a lack of resources. For example, caregivers and HCPs reported that transitioning from paediatric to adult healthcare was difficult for autistic individuals (69). Continuity of care can be challenging and complex due to the various comorbidities that can exist alongside autism, necessitating care across a number of services. However, there is an identified lack of support in planning for, or making, transitions (101). The rigidity of the healthcare system also caused problems for all three groups (i.e., autistic individuals, caregivers, and HCPs). For example, HCPs reported that a lack of time, resources, support, and guidelines prevented them from making necessary accommodations (51, 64). Similar findings were reported by Tuffrey-Wijne and colleagues (84) who found that a lack of clear lines of responsibility, a lack of funding and staffing, and not knowing what adjustments were needed, were reported as barriers by healthcare staff to making reasonable adjustments to healthcare for persons with intellectual disabilities in the UK. Table 2 provides examples of potential solutions to issues within this theme. For example, developing a shared plan of care, involving the patient and/or the caregivers and the HCPs may facilitate easier transitions through the care continuum. For example, Wittling et al. (102) found that behavioural coping plans, developed through collaboration between HCPs and caregivers, allowed for individualised interventions to be implemented throughout the care process.

Other Patient-related factors

The final theme encompassed additional challenges associated at the patient level. Caregivers' attitudes towards, or engagement with, complementary alternative medicine was seen as a barrier by some HCPs. Families often engage in a wide variety of 'treatments' for autism, many of which are not evidenced-based (103, 104). Green and colleagues (105) found that families were currently using an average of seven treatments for their autistic child, and that whether or not a treatment was evidenced-based did not seem to influence the preference for use of that treatment. With the availability and accessibility of information via the internet, families are exposed to recommendations for a vast array of treatments, with varying degrees of evidence, which they may in turn suggest to their HCPs (106). This was reflected in the current study as caregivers often reported that their HCP's reluctance to discuss alternative treatments could be a barrier to care. Green and colleagues (105) recommend that objective, data-based, consumer-friendly information regarding evidence-based interventions for autism should be readily accessible to parents in order to counter the use of pseudoscientific treatments. HCPs are well placed to provide this information; however, a number of studies have shown that HCPs are not sufficiently knowledgeable about such treatments (93, 107) and that families of autistic children were more likely to report that physicians were less knowledgeable about complementary alternative treatments than families of children with physical disabilities or intellectual disability (11). Table 2 highlights the use of a training program that was shown to increase physicians' knowledge of evidence-based treatments. However, future research should also aim to increase knowledge of non-evidenced based treatments and intervention so that HCPs can advise and guide families appropriately.

Strengths and limitations

This systematic review has a number of strengths which may be noted. A rigorous, systematic search strategy was applied across five databases and the methodological quality of the included studies has been assessed in a systematic manner using a standardised set of criteria. Additionally, the perspectives of three stakeholder groups have been examined and a consistent set of challenges was observed. This triangulation of the data from the perspectives of three different stakeholder groups has enabled a more robust and comprehensive understanding of the reasons that autistic individuals experience access problems in healthcare. Finally, the validity of the findings is strengthened by the convergence of data from the various sources into coherent themes (108).

It is important to note that this review also had a number of limitations. First, the review only included articles that have been published in peer-reviewed journals as the inclusion of grey literature can create issues due to: 1) the lack of best practice guidance on how and where to conduct searches; 2) the difficulty in interpreting data due to potentially low methodological quality and poor reporting; and 3) the difficulty in reproducing searches (109-111). Nonetheless, this may lead to a risk of publication bias. Second, due to resource constraints, it was only possible to include studies that were published in English and for one author to screen titles and abstracts at the screening stage. However, whenever there was uncertainty about the eligibility of an article, it was discussed with the other members of the research team until consensus was reached. A third limitation is that the review focused solely on barriers to healthcare. It is possible that the inclusion of facilitators, that were reported in a number of the included studies (e.g., consistency and familiarity in the healthcare environment (49); caregivers being involved in the planning of the healthcare visit; 60), would have provided greater insight into the healthcare experiences of autistic individuals and how best to improve care. However, the taxonomy that has been produced has clear and direct implications for evaluation of services and quality improvement and inclusion of facilitators may have detracted from this. Future researchers may wish to consider the research literature on facilitators and how this data may also contribute to quality improvement. Finally, analysis relied on barriers as presented by the authors of the included studies, without any inference. In some cases, issues attributed to the HCP (e.g., appointments being too short) are likely to be systemic issues that autistic individuals or caregivers may not recognise. However, the data provided within the included papers did not allow for this kind of analysis or inference.

Future research

The data collated in this review allow for a number of recommendations to be made for future research and practice. First, it is worth noting that autistic individuals participated in just seven of 30 studies in this review (23%), whereas caregivers were included in 16 studies (53.33%) and HCPs were included in 14 studies (46.66%). This is consistent with Mason and colleagues (29) recent systematic review of barriers to physical healthcare for autistic adults which found just six articles where adults on the autism spectrum were participants. This reflects a lack of input and contribution from the group whom this issue affects the most. It is becoming increasingly accepted and recommended that autistic individuals should be involved in, and consulted about, autism-related research (41,73, 112). Therefore, it is imperative that the perspectives of autistic individuals are taken into account in future research and work in this area, so that their healthcare needs are better understood, and high quality, evidence-based models of care can be developed (113). It was not possible to compare barriers reported by autistic adults and children as the majority of studies representing children rely on caregiver report. In the two studies where children were included as participants, however, there do seem to be some similarities in barriers experienced (e.g., communication difficulties, sensory sensitivities) (54). Further research is required to fully explore the differences and similarities in barriers experienced by adults and children on the spectrum in order to tailor interventions effectively.

It is notable that the majority of studies were published since 2015, indicating a growing amount of research aimed at improving the health and provision of care for people on the autism spectrum. There are likely a number of reasons for this, such as the expressions of desire from the autism community and their supporters for research which focuses on improving day-to-day life (114), the governmental Acts in the UK (e.g., the Autism Act, 2009) which stipulate that public services need to adjust their care to accommodate people with disabilities, and the introduction of funding specifically for this type of research from organisations such as Autistica UK (115). However, other researchers or literature reviews could consider other reasons for this increased focus in more detail.

It is hoped that in future research, this taxonomy can be used to facilitate the development of measures that can be employed in service evaluation, when identifying adaptations required by an individual, or to allow for benchmarking across services. In the studies reviewed, a variety of measures were used to assess the barriers experienced by autistic individuals, caregivers, and HCPs in healthcare contexts. The findings from our analysis, however, indicate that there does not appear to be a single measure that can provide a comprehensive assessment of all the barriers that are experienced at the various levels by the different stakeholders. For example, although Raymaker et al. (41) developed a valuable tool for assessing barriers from the perspectives of autistic individuals, it is not exhaustive, as a number of barriers that are reported by autistic individuals in other studies were not accounted for, such as stigma (e.g., 57,70) or struggling with the unpredictability of healthcare settings (e.g., 44). Golnick et al. (71), Carbone et al. (81) and Will et al. (65) used a measure of nine items relating to barriers from the perspective of HCPs. These included barriers such as a lack of reimbursement and families being sceptical about vaccines but did not account for issues such as difficulties communicating with the patient. Similarly, Ahmedani & Hock (45) surveyed caregivers on items such as financial issues and not feeling like a partner in their child’s care but did not account for negative perceptions (e.g., 64) or a lack of provider knowledge and skill (e.g., 54). Additionally, many of the measures used targeted only one participant group (e.g., Chiri & Warfield (46) focused solely on caregivers). The development of a measure that could be used to conduct a comprehensive audit of healthcare settings, considering the perspectives of the three stakeholder groups at the patient, provider, and system levels, would afford a more holistic overview of the accessibility of healthcare settings for autistic individuals and identify the accommodations required. It is also recommended that future research explore the barriers experienced in specific healthcare settings, such as office- or hospital-based settings in order to assess whether there are unique difficulties depending on the specific context.

Finally, few studies have examined the links between the barriers experienced and health outcomes of autistic individuals. Many of the included studies examined factors such the type of access barriers experienced (e.g., 44), HCP self-efficacy (e.g., 81), HCP knowledge of autism (e.g., 51), and satisfaction with care (e.g., 46). In other areas of healthcare research, improved satisfaction with primary care has been shown to be associated with continuity of care (116) and patient compliance with treatment regimens (117), that may positively affect patient outcomes. It would, therefore, be useful to determine if an intervention targeting barriers and any observed improvements in factors such as satisfaction with care or autism knowledge translate into better health for autistic individuals. Future research should examine this in order to facilitate an understanding of which barriers are most important with regards to health outcomes and should, therefore, be targeted by intervention with the limited resources available.

Conclusion

As the prevalence of autism among both adults and children increases, the availability of appropriate HCPs and effective healthcare services for autistic individuals becomes ever more essential. The taxonomy developed herein offers a clear and useful overview of the various challenges and barriers that can impede the delivery of effective care for autistic individuals in healthcare settings. This taxonomy has clear and important implications for interventions to improve access to healthcare for autistic individuals and the quality of healthcare that is received. Such initiatives may contribute towards improving the health outcomes and healthcare experiences of autistic individuals.

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Chapter 3: Study 2

Development and Evaluation of a Novel Caregiver-Report Tool to Assess Barriers to Healthcare for People on the Autism Spectrum

Development and evaluation of a novel caregiver-report tool to assess barriers to healthcare for people on the autism spectrum

Where this fits in with the thesis

Adequate standardised caregiver-report tools are important as not all autistic individuals can self-report due to factors such as age, communication abilities or co-occurring intellectual disabilities. Much of the research on healthcare and autism has relied on caregiver-report (1, 2). However, Study 1 (Chapter 2) indicated that there is a lack of standardised caregiver-report measurement tools which can assess barriers to healthcare that occur across the healthcare system, and that are informed by barriers identified by autistic individuals. Chapter 3, therefore aimed to develop a caregiver report tool to address the research question: *what are the barriers to physical healthcare reported by caregivers of autistic individuals living in Ireland?* It is hope that this tool will facilitate the identification of barriers to healthcare experienced by autistic individuals and facilitate conversations between healthcare providers, caregivers, and patients about challenges experienced and required adjustments.

Peer-reviewed publication

This study has been accepted and published in a peer-reviewed journal.

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The following chapter is a formatted version of the submitted manuscript to the journal.

Conference presentations

Oral presentation

Walsh C, Lydon S, Hehir A, O'Connor P. Development and evaluation of a novel caregiver-report tool to assess barriers to physical healthcare for people on the autism spectrum. Paper presented at the Health Services Research UK Conference 2021. July 6th 2021; Virtual event

Poster presentation

Walsh C, Lydon S, Hehir A, O'Connor P. Development and evaluation of a novel caregiver-report tool to assess barriers to physical healthcare for people on the autism spectrum. Poster presented at the Trinity Health and Education International Research Conference 2021; March 9th-11th, 2021. Virtual event.

Author contributions

CW, POC and SL proposed and designed the study. CW and AH collected the data. CW analysed the data and drafted the manuscript. CW, SL, and POC contributed to the manuscript. All authors have read, commented on, and approved the final manuscript.

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Abstract

Introduction: People on the autism spectrum often experience poorer health than the general population despite higher engagement with the health services. This suggests a disparity in the accessibility of appropriate healthcare for autistic individuals. To improve access, barriers the autism community experience in healthcare first need to be identified. This paper aimed to: 1) develop and evaluate a caregiver-report tool; 2) identify barriers to physical healthcare for autistic individuals; and 3) identify potential contributing factors.

Methods: A previously established taxonomy of barriers to healthcare for autistic individuals informed the development of the tool; this was then distributed to caregivers of autistic adults and children. Exploratory factor analysis (EFA) assessed validity and reliability of the tool. Multiple regressions were performed to identify predictors of barriers.

Results: In total, 194 caregivers participated in the study. The EFA produced four factors: 1) patient-related barriers; 2) healthcare provider (HCP)-related barriers; 3) healthcare system-related barriers; and 4) barriers related to managing healthcare. The most commonly endorsed barriers included difficulties with identifying/reporting symptoms (endorsed by 62.4% of participants); difficulties handling the waiting area (60.3% of participants); and a lack of HCP knowledge regarding autism (52.1% of participants). Autism severity, general adjustment problems, anxiety, age and having unmet needs predicted the frequency and/or severity of barriers.

Conclusions: A tool that allows assessment of patient-, HCP-, and system-level barriers to healthcare was developed and evaluated. Patient-level barriers appear to occur frequently and pose substantial challenges. This tool will help identify areas most in need of intervention and support intervention evaluation.

Keywords: Autism Spectrum Disorder; Healthcare Access; Reasonable Adjustments; Healthcare Equity; Healthcare Disparities; Health;

Conflict of interest

The authors declare they have no conflicts of interest.

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Introduction

Individuals on the autism spectrum may require a greater degree of interaction with health services due to the multitude of medical comorbidities that often present alongside autism (e.g., gastrointestinal issues, psychiatric conditions, intellectual disabilities; (1-3). Yet, despite the higher rates of contact with both emergency and non-emergency healthcare (4, 5) autistic[2] individuals tend to have poorer health outcomes (2, 7), engage less with preventative care (8, 9), have lower health-related quality of life (10) and higher mortality rates (11) than others. This suggests that autistic individuals face significant inequities in healthcare.

A number of determinants of health inequities for people with learning and developmental disabilities have been outlined (12). These include social determinants (e.g., poverty, unemployment); poorer communication and health literacy; poor health behaviours; and deficiencies in access to, and quality of, healthcare. Although research suggests that autistic individuals share many of these determinants of health inequities, this particular population may face additional, unique challenges as the lack of a ‘visible disability’ may lead to a misperception that they experience less barriers and therefore require less support in healthcare settings (9, 13).

Improving access to healthcare may be one way to improve health equity for autistic individuals. Healthcare access is a complex issue for which a number of conceptualisations exist (14). According to Penchansky and Thomas (15), access should be viewed as the ‘fit’ between the patient’s needs and the health service’s ability to meet those needs. This requires consideration of the availability, accessibility, accommodation,

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[2] Many autistic individuals and their supporters prefer the use of identity-first language (i.e., autistic person) as opposed to person first language (i.e., person with autism; 6). Accordingly, this chapter uses identity first language.

affordability, and acceptability of healthcare services. Shengelia, Murray and Adams (16) conceptualised access as the likelihood of receiving a healthcare service when it is needed (coverage), and the quantity of healthcare services and procedures used (utilisation). Similarly, Levesque, Harris, & Russell (17) describe access as the ability to identify healthcare needs, seek services, reach resources, obtain, or use services, and be offered services appropriate to needs. Although various conceptualisations of access exist, there appears to be a fundamental commonality that access requires both getting into a service and receiving the appropriate and necessary care, when it is needed.

Disparities in access to healthcare can lead to delays in, or non-receipt of, appropriate care (18); higher financial burden on the patient (19); increased use of emergency care (20); higher hospitalisation rates (21); higher risk for, and poor management of, chronic illness or comorbid conditions (22): greater risk of unmet needs (23), and increased burden and cost on the healthcare system due to preventable disease exacerbations and premature deaths (24). Ready access to healthcare on the other hand, is linked to a variety of important outcomes including better health status (9), higher patient satisfaction (25), lower hospitalisation rates (26), and reduced burden and cost on the healthcare system (27). Given the benefits of improved healthcare access to both the health services and the patient, it is important that access issues are examined and remedied among the groups for whom access is impaired.

A growing body of research has identified a complex array of access barriers for autistic people. In a previous taxonomy (28), barriers to physical healthcare which were reported/endorsed by autistic individuals, caregivers, and HCPs, were categorised as occurring at the level of the patient (e.g., difficulties with social interactions and communication (29); the HCP (e.g., a lack of physician knowledge about autism (30); and the healthcare system (e.g., a lack of support for autistic individuals and their families; 31). Other literature has observed similar results (32, 33).

Addressing these complex barriers to accessing healthcare requires an understanding of the specific issues that impact on the ability of patients to access the appropriate care in their services. It is therefore important that measurement tools designed to assess barriers to accessing healthcare consider barriers across the healthcare system. A recent systematic review (28) indicated that few caregiver-report tools consider the barriers across the healthcare system (i.e., patient, HCP, and systems levels). Although Raymaker et al. (34) have developed a thorough self-report tool for autistic adults, not all autistic individuals can self-report on their experiences due to factors such as age or comorbid intellectual disabilities. Therefore, it is important to provide caregiver-report tools that ensure good measurement of potentially diverse barriers. Such tools will enable the identification and evaluation of initiatives that aim to reduce barriers and improve healthcare access or experiences.

Study aims

The aims of this study were to: 1) develop a valid caregiver-report tool to measure barriers to healthcare for autistic individuals; 2) use the newly developed tool to examine the barriers to care endorsed by caregivers; and 3) identify potential contributing factors such as autism severity, comorbid psychopathologies, or having unmet needs.

Methods

Participants

Eligible participants were: 1) the primary caregivers to an autistic child or adult; 2) over the age of 18; and 3) living in the Republic of Ireland.

Measures

Survey instrument

The survey instrument administered to participants consisted of six sections: 1) demographics; 2) barriers to healthcare; 3) frequency of contact with the health services; 4) perceived unmet healthcare needs; 5) autism severity; and 6) presence of co-occurring psychopathology.

Demographics

Caregivers were asked to provide information on both themselves (i.e., age, gender, location, and highest level of education, relationship to the autistic person), and the autistic individual for whom they care (i.e., age, gender, health status).

Barriers to Healthcare tool

Literature review

A systematic literature review on barriers to healthcare was conducted and is described in detail elsewhere (28). Individual barriers reported by autistic adults, caregivers and HCPs in the included studies were categorised into a taxonomy of barriers containing 4 themes (autism-related characteristics; other patient-related barriers; HCP-level barriers; system-level barriers). These themes informed the item construction of the tool.

Item construction

An iterative method was used to construct the items of the questionnaire, following best practice (35). This involved two consensus building meetings between three members of the research team (CW, SL, & POC). The specific items from existing questionnaires were deliberately not reviewed as part of this process as the team wanted to work from the themes within the previous taxonomy (28). It is acknowledged, however, that because there are a finite number of ways to ask about a particular barrier, similarities with items in existing measures may arise through convergence as part of the development process. In the first meeting, questionnaire items (*n*=57) were constructed to address the four themes in the taxonomy. During the subsequent meeting, these items were reviewed, refined, and condensed into 42 items, each representing a different barrier. Respondents were asked to indicate: 1) the *frequency* with which that barrier had occurred in the past 12 months on a Likert scale ranging from 0 (never) to 4 (very often); and 2) the *severity* with which that barrier was experienced on a Likert scale of 1 (slight), 2 (moderate), and 3 (severe). The response options were adapted from the Behaviour Problems Inventory (36).

Frequency of contact with the healthcare services

To indicate the frequency of contact with the healthcare services, participants provided information in response to the following three items: 1) number of general practice (GP) visits in the past 12 months; 2) number of emergency department (ED) visits in the past 12 months; and 3) number of hospital visits in the past 12 months.

Unmet needs

Participants were asked to indicate whether unmet healthcare needs had occurred in the last 12 months in relation to GP, hospital, emergency care, mental healthcare service, or ‘other’ services. This item was adapted from Nicolaidis et al. (9). For the purpose of analysis, this variable was collapsed into two categories, (i.e., whether they had experienced an unmet need, or not).

Autism severity

The Social Responsiveness Scale-2 (SRS-2) was included in the questionnaire as a measure of autism severity (37). This is a 65-item validated scale that has previously demonstrated high internal consistency

(Cronbach's alpha=.95; 38). There are four versions of the SRS which are intended to allow autism severity to be measured across all age groups: 1) caregiver report: pre-school children, 2.5-4.5 years; 2) caregiver report: school age children, 4.5-18 years; 3) caregiver report: autistic adults, 18+ years; 4) autistic adults self-report. Across all versions, the items, which describe the autistic person's behaviour over the past 6 months, are similar but worded as appropriate for the respondent (i.e., caregiver/self-report) and developmental stage (child/adult). Each item is scored on a Likert scale ranging from 0 (not true) to 4 (almost always true).

Anxiety, Depression, Obsessive Compulsive Disorder, General Adjustment Problems

The Psychopathology in Autism Checklist (39) was used as a measure of comorbid psychopathological issues that may be experienced by autistic individuals. The tool consists of 42 items which correspond to one of four psychiatric disorders: Anxiety (6 items), Depression (7 items), Obsessive Compulsive Disorder (OCD; 7 items), Psychosis (10 items); and General Adjustment Problems (GAP; e.g., sleep disturbances, self-harm, challenging behaviour; 12 items). Four subscales (Anxiety, Depression, OCD, General Adjustment Problems) which have previously demonstrated good internal consistency (39) were used in the current analysis. Each item is rated on a Likert scale ranging from 1 (not a problem) to 4 (severe problem). The average score per item per subscale is used in analysis.

Procedures

Recruitment and data collection

A variety of non-probability sampling methods and recruitment strategies were employed in an effort to recruit as wide a sample as possible including: 1) sending letters of invitation with a recruitment flyer to primary and secondary schools (*n*=395) which had an ASD unit or special needs class, and to special schools (*n*=80) in the Republic of Ireland. Principals were asked to disseminate information on the study and/or distribute questionnaires to the parents of their students; 2) autism parent support groups and autism and intellectual disability support services/organisations were contacted and asked to share information about the study and/or distribute questionnaire packs to their members; and 3) advertisements were placed in local and national newspapers, on local radio, and were posted on social media. Snowballing methods were also employed whereby participants were asked to share information about the study with others. Potential participants were asked to contact the primary author to access further information about the study or to request a survey pack which was either sent to the participant by post with a stamped return envelope, or could be accessed online. Participants were offered the opportunity to enter a prize draw to win one of four €50 shopping vouchers as an incentive to participate.

Ethical approval

Ethical approval for this study was granted by the NUIG Research Ethics Committee (Ref: 17-Nov-20). Informed written or electronic consent was obtained for each participant.

Statistical analysis

All analyses were performed using IBM SPSS Statistics 24. Significance levels for all analyses were set as p<.05.

Initial data screening

Little's test for Missing Completely at Random was conducted to ensure the missing data were randomly distributed. Missing Data Analysis in SPSS was also used to assess the missing data. Preliminary analyses were performed to ensure no violations of the assumptions of normality, multicollinearity, and homoscedasticity. The reliability of the predictor variable scales (Anxiety; Depression; OCD; GAP; Autism Severity) was assessed by calculating Cronbach’s alphas.

Addressing aim 1: Development of a valid and reliable caregiver-report Barriers to Healthcare tool

Construct validity

Construct validity is concerned with whether the items in the subscale reflect the same construct (40). A way to assess this assertion is to use exploratory factor analysis (EFA; 41). The approach to EFA outlined by Costello and Osborne (42) was undertaken.

Step 1: Adequacy of the correlation matrix.
The suitability of the data for use within an EFA was assessed by considering the sample size, factorability of the constructs (correlation matrix), examination of the Kaiser-Meyer-Olkin (KMO) Measure of Sampling Adequacy and Bartlett’s Test for Sphericity.

Step 2. Factor extraction, retention, and interpretation.
Principal Axis Factoring was chosen as the factor extraction method as this focuses explicitly on latent factors (42). Factor extraction was determined by considering Kaiser’s criteria (Eigenvalue >1), the scree plot, and a parallel analysis (PA; via an online PA engine; 43). Oblique (Promax) rotation was used as the data cannot be assumed to be completely independent of each other. This is considered most accurate for research involving human participants (42). Items with factor loadings of greater than 0.4 was chosen as a cut off for factor retention in the current dataset (44). The pattern matrix guided interpretation and naming of the factors by the research team (44, 45).

Step 3. Internal consistency.

Internal consistency (the extent to which items within a construct are inter-correlated; 46) was assessed by calculating Cronbach’s alpha for each construct produced by the EFA.

Addressing aim 2. Examine the barriers to care endorsed by caregivers

Examining barriers endorsed by respondents in terms of frequency

A total Frequency of Barriers scale score was calculated by summing all items that remained after the EFA. Subscale scores were then calculated for each of the extracted factors after the EFA.

Examining the barriers endorsed by respondents in terms of severity

A total Severity of Barriers scale score was calculated by summing all items. No subscale scores were calculated as no EFA was conducted due to insufficient data.

Addressing aim 3: Identify related variables

Identifying variables related to frequency of barriers

A series of five hierarchical multiple regressions was performed to assess whether the frequency of barriers was associated with the following variables: 1) age; 2) gender; 3) health status; 4) perceived unmet needs; 5) Autism Severity; 6) Anxiety; 7) Depression; 8) OCD; or 9) General Adjustment Problems. Frequency of contact with the healthcare services was controlled for because it is likely that individuals who have a higher frequency of contact with the healthcare services are more likely to report a higher frequency of barriers. The same method was used for each of the five regressions with just the criterion variable changing each time. These variables were selected on the basis of a systematic review examining barriers to healthcare (28).

Step 1.
In each regression, frequency of contact with the health services (i.e., number of GP, ED, and hospital visits in the past 12 months) were controlled for by entering them in the first step.

Step 2.
Age, gender, and health status (good/poor) of the autistic individual; perceived unmet healthcare needs (yes/no); Depression; Anxiety; OCD; General Adjustment Problems; and Autism Severity were all entered in the second step. The five criterion variables were: 1) the overall frequency of barriers endorsed; 2) the frequency of barriers endorsed at the patient level; 3) the frequency of barriers endorsed at the healthcare provider level; 4) the frequency of barriers endorsed at the systems level; and 5) the frequency of barriers endorsed relating to managing healthcare.

Predicting severity of barriers

A multiple regression was performed to assess the predictive values of all predictor variables on the perceived severity of barriers. In this analysis, the criterion variable was the total score for perceived severity of barriers and the predictor variables were: age, gender, health status (good/poor), Autism Severity; unmet need (yes/no), contact with health services (i.e., number of GP, ED, and hospital visits in the past 12 months), Anxiety; Depression; OCD; and General Adjustment Problems.

Results

Response rate

Due to the various recruitment methods (i.e., advertising and distributing information leaflets), it is not possible to provide an accurate response rate. A total of 403 surveys were sent, and 194 were returned – an estimated response rate of 48.14%.

Participants

Caregivers of 194 autistic children/adults participated in the study. Descriptive statistics indicated that the mean age of caregiver respondents was 42.26 years (*SD*=8.15, range 23-63 years). Respondents were mostly female (*n*=162; 83%) and were mostly mothers of the autistic individual (*n*=145, 74.7%). The majority of autistic individuals about whom the survey was completed were male (*n*=155; 79.9%) and had a mean age of 12.47 years (*SD*=8.89, range: 2-64 years). Table 1 provides more detail on sample characteristics.

Table 2 presents information on participant scores for frequency and severity of barriers, and scores on all predictor variable scales (i.e., scores on the subscales of the Psychopathology in Autism Checklist: Anxiety, Depression, OCD, & GAP; scores on the SRS-2: Autism Severity; scores on perceived unmet needs and the contact with the health services variables).

Table 1. Participant characteristics

Caregiver Characteristics	***N*(%)**
Age: M, (SD), Range	
42.26 (8.15); 23-63years	
Gender	
Female	162(83.1)
Male	33(16.9)
Education	
Primary	2(1)
Secondary	41(21)
Third level (Undergraduate)	66(33.8)
Third level (Postgraduate)	68(34.9)
Diploma/certificate/trade	17(8.76)
Location	
Rural	73(37.6)
Village	25(12.9)
Town	67(34.5)
City	29(14.9)
Relationship to autistic individual	
Mother	145(74.7)
Father	24(12.4)
Other relative	1(.5)
Care worker	24(12.4)
Characteristics of Autistic Individuals (*N*=194)	
Age M (SD); Range	
12.47 years (8.89); 2-64 years	
<10	92(47.4)
10-20 years	74(38.1)
21-30 years	18(9.3)
31-40 years	6(3.1)
>40 years	4(2.1)
Gender	
Female	39(20.1)
Male	155(79.9)
Health status	
Excellent	74(38.1)
Good	96(49.5)
Fair	20(10.3)
Poor	4(2.1)
Unmet need during past 12 months	
GP	22(11.3)
Routine hospital care	57(29.4)
Emergency care	10(5.2)
Mental healthcare	77(39.7)
Other	53(27.3)
Contact with health services during past 12 months	***M* (SD); Range**
GP	3.9(4.12); 0-24
Routine hospital care	2.97(5.07);0-41
Emergency care	0.52(1.29); 0-12

GP=general practice

Table 2. Participant scores on barriers scales and predictor variables scales

Scores on Barriers Scales	***N***	**M(*SD*)**	**Range**	**Scores on Predictor Variable Scales**	***N***	**M(*SD*)**	**Range**	**≥ cut-off point *N*(%)**
Total frequency of barriers;	194	54.8 (23.06)	5-119	Depression	187	1.94 (.60)	1-3.71	n/a
Frequency of patient-related barriers;	194	23.63 (8.94)	1-40	Anxiety	187	1.69 (.56)	1-3.83	n/a
Frequency of HCP-related barriers;	194	12.16 (6.28)	0-24	OCD	187	1.78 (.61)	1-3.57	n/a
Frequency of system-related barriers;	194	14.02 (9.90)	0-40	General Adjustment Problems	187	2.26 (.57)	1-3.92	n/a
Frequency of barriers related to managing healthcare;	194	5.27 (3.78)	0-16	Autism Severity	187	108.61 (26.44)	50-182	171 (91.44)
Total severity of barriers;	121	37.45 (15.95.)	5-80					

HCP=healthcare provider; OCD=Obsessive Compulsive Disorder

Aim 1. Development of a valid and reliable and caregiver-report Barriers to Healthcare tool

Initial data screening

All assessments of missing values indicated that the missing data were randomly distributed, with no item incurring >30% missing data. Therefore, simple mean imputation was used to replace the missing values (47). No items were skewed (i.e., had a value greater/less than 3/-3). Three items showed some level of kurtosis (i.e., greater/less than 3/-3; range 3.2-4.1). Since this was an exploratory procedure, these variables were retained and monitored to see if they were having any undue influence on analysis.

Construct validity

Step 1. Suitability of the data to an EFA.

No pairs of items were highly correlated (>0.8), suggesting that the correlation matrix was adequate, and multicollinearity was unlikely to be an issue (45). Bartlett's test of sphericity indicated the correlation matrix was not an identity matrix (χ^2=4389.062, *df*=861, *p*<.001). The Kaiser-Meyer-Olkin (KMO) measure of sampling adequacy also indicated that the matrix was suitable for EFA (KMO=0.89). Examination of the Measures of Sampling Adequacy (MSAs) along the principal diagonal of the anti-image correlation matrix indicated that all variables were suitable for inclusion in the EFA as all had a value greater than 0.8 and off diagonal values were small (<0.3; 48).

Step 2. Factor extraction, retention, and interpretation.

All 194 participants were included in the EFA. In the current dataset the Eigenvalues >1 rule extracted 10 factors; however, this method has a tendency to over extract factors and so is not generally recommended (48). The scree plot suggested retaining three factors. However, since the sample size was <200, it is not advisable to rely on the Scree plot alone (44). To determine the number of factors more rigorously, Parallel Analysis (PA) (49) was conducted. The PA suggested retaining four factors as the Eigenvalues for the first four components exceeded the corresponding Eigenvalues generated by random data at the 95th percentile (Appendix 2.1). Correlations between the factors were observed in the factor matrix suggesting an oblique rotation (Promax) be maintained (45). The four-factor solution cumulatively explained 50.86% of the variance. Nine items did not meet the loading criterion of $\geq$0.4 (44). so were discarded from the model.

On the next iteration all items loaded above 0.4. However, three items loaded with a difference of <2 onto more than one factor. The EFA was run again with these items removed. On the next iteration, all items had a factor loading of $\geq$0.4; one item cross loaded onto two variables with a difference of less than 2, so was discarded. On the next iteration, another item did not have a loading of $\geq$ 0.4 on any factor, so was deleted. In the following iteration one item cross loaded onto two factors with a difference of less than 2, so was discarded. On the final iteration, all remaining items had factor loadings greater than 0.4; any evident cross loadings showed a greater difference than 2. This final model explained 50.82% of the variance. Table 3 presents the four extracted factors with the corresponding items, factor loadings, and the amount of variance explained by each factor. The items which had been observed as Kurtosed earlier were discarded during the EFA process (Appendix 2.2 for item deletion process). The final Barriers to Healthcare tool contained 27 items and requires approximately 10 minutes to complete (Appendix 2.3).

Step 3. Internal consistency

As shown in Table 3, Cronbach's alphas indicated that all four factors showed good internal consistency as per conventional standards of interpretation (50).

Table 3. Factor loadings after EFA, internal consistency scores, and variance explained

Item	Factor 1	Factor 2	Factor 3	Factor 4
Factor 1: Patient-related barriers (Cronbach's α=.889; 32.9% variance explained)				
Child/adult finds appointments overwhelming;	.841			
Child/adult finds it difficult to tolerate medical procedures;	.725			
Child/adult finds it difficult to handle the waiting area;	.697			
Child/adult dislikes doctors;	.674			
Healthcare facilities cause child/adult sensory discomfort;	.624			
Caregiver worries that stress of interacting with the healthcare system will cause child/adult distress;	.601			
Caregiver is afraid to take child to the doctor;	.586			
Caregiver finds healthcare appointments overwhelming;	.584			
Child/adult has difficulty identifying/reporting pain and other symptoms;	.582			
Child/adult has difficulty communicating their thoughts, wants or needs;	.537			
Factor 2: System-related barriers (Cronbach's α=.865; 8.86% variance explained)				
Insurance impacts access to healthcare;		.825		
Cost of care impacts access to healthcare;		.759		
Insurance does not cover the care we need;		.727		
Transport costs too much;		.692		
There are other transportation problems;		.562		
Inadequate caregiver/family support;		.513		
Caregiver does not know where to go;		.454		
Factor 3: HCP-related barriers (Cronbach's α=.876; 5.07% variance explained)				
HCPs do not listen;			.825	
Child's/adult's behaviours are misinterpreted by HCPs;			.810	
HCPs have inadequate knowledge/education for treating autistic people;			.722	
Caregiver does not like how HCPs have treated child/adult in the past;			.705	
Finding a HCP who will accommodate child's/adult's needs can be difficult;			.635	
I know others who have had negative healthcare encounters;			.614	
Factor 4: Barriers related to managing healthcare (Cronbach's α=.753; 4.03% variance explained)				
Medical recommendations given to child/adult can be hard to follow;				.697
The treatment plan given to child/adult can be hard to follow;				.582
Caregiver finds it difficult to remember to attend appointments;				.556
Following up on child's/adult's care can be difficult;				.530

HCP=healthcare provider

Aim 2: Examine the barriers to care endorsed by caregivers

Frequency scores

All 194 participants were included in this analysis. Regarding patient-related barriers, more than half of respondents indicated that the autistic individual’s difficulty identifying/reporting pain/symptoms (62.4%), finding it hard to handle the waiting area (60.3%) and being unable to communicate thoughts, wants or needs (53.1%) occurred ‘often’ or ‘very often’. Regarding HCP-related barriers, a lack of HCP knowledge was reported by over half of respondents (52.1%) as occurring ‘often’ or ’very often’. At the systems level, almost half of respondents (46%) indicated that a lack of support was occurred ‘often’ of ‘very often’. For full details of barriers endorsed, see Table 4.

Severity scores

The missing data were found to be randomly distributed for the Severity of Barriers scale. However, a large number of items had a relatively high percentage of missing data (range: 4.6%-11.9%). On closer inspection, it was inferred that data were missing due to misinterpretation of the scale due to its presentation in the questionnaire (Appendix 2.3). Participants were asked to choose two responses for each item to indicate: 1) frequency, and 2) severity. However, many participants responded to the frequency scale alone. Therefore, simple mean imputation was not deemed suitable, listwise deletion was used instead. Cronbach’s alpha indicated good internal consistency (α=.925).

Difficulty identifying/reporting pain/symptoms was endorsed as severe by 40.2% (*n*=78) of respondents. This was followed by the autistic individual not being able to communicate their thoughts, wants or needs (36.1%; *n*=70), and a lack of HCP knowledge, which was endorsed as severe by 33% (*n*=64) of respondents. See Table 4 for further details. As listwise deletion was used, the sample size for each item varies, the percentages provided reflect the number of participants who responded to that item.

Table 4. Endorsement rates of barriers as 'often' or 'very often' and 'severe'

	Often/very often *N* (%)	**Severe *N* (%)**
1. Patient-related barriers		
Child/adult finds appointments overwhelming;	79 (40.7)	40 (20.6)
Child/adult finds it difficult to tolerate medical procedures;	95 (49.0)	39 (20.1)
Child/adult finds is difficult to handle the waiting area;	117 (60.3)	69 (35.6)
Child/adult dislikes doctors;	61 (31.4)	35 (18.0)
Healthcare facilities cause child/adult sensory discomfort;	87 (44.8)	50 (25.8)
Caregiver worries that the stress of interacting with the healthcare system will cause child/adult distress;	92 (47.4)	39 (20.1)
Caregiver is afraid to take child/adult to the doctor;	36 (18.5)	16 (8.2)
Caregiver finds healthcare appointments overwhelming;	59 (30.4)	25 (12.9)
Child/adult has difficulty identifying/reporting pain or other symptoms;	121 (62.4)	78 (40.2)
Child/adult has difficulty communicating their thoughts, wants or needs;	103 (53.1)	70 (36.1)
2. System-related barriers		
Insurance impacts access to healthcare;	65 (33.5)	38 (19.6)
Cost of care impacts access to healthcare;	66 (34)	38 (19.6)
Insurance does not cover the care we need;	67 (34.5)	43 (22.2)
Transport costs too much;	41 (21.2)	17 (8.8)
There are other transportation problems;	24 (12.4)	12 (6.2)
Inadequate caregiver/family support;	90 (46.4)	53 (27.3)
Caregiver does not know where to go;	21 (10.8)	10 (5.2)
3. HCP-related barriers		
HCPs do not listen;	56 (28.8)	31 (16.0)
Child's/adult's behaviours are misinterpreted by HCPs;	63 (32.5)	29 (14.9)
HCPs have inadequate knowledge/education for treating autistic people;	101 (52.1)	64 (33.0)
Caregiver does not like how HCPs have treated child/adult in the past;	45 (23.2)	31 (16.0)
Finding a HCP who will accommodate child's/adult's needs can be difficult;	77 (39.7)	39 (20.1)
Caregiver knows others who have had negative healthcare encounters;	89 (45.9)	48 (24.7)
4. Barriers related to managing healthcare		
Medical recommendations given to child/adult can be hard to follow;	42 (21.6)	15 (7.7)
The treatment plan given to child/adult can be hard to follow;	41 (21.2)	18 (9.3)
Caregiver finds it difficult to remember to attend appointments;	23 (11.8)	10 (5.2)
Following up on child's/adult's care can be difficult;	45 (23.2)	11 (5.7)

HCP=healthcare provider

Aim 3: Identify variables associated with the frequency of barriers endorsed (total barriers, patient-related barriers, HCP-related barriers, systems-related barriers, barriers related to managing care)

Initial data screening

Missing data were found to be randomly distributed. However, seven cases were missing more than 30% of the data so were removed from the regression analyses. Simple mean imputation was used for the remaining cases (*n*=187). No violations of the assumptions of normality, multicollinearity and homoscedasticity were observed (Appendix 2.4). All predictor variable scales indicated good internal consistency in the current study, as per conventional standards (50; SRS α=.923; Anxiety α=.756; Depression α=.754; OCD α=.796; General Adjustment Disorder α=.816)

Variables associated with the frequency of barriers

Total frequency of barriers.

As shown in Table 5, Autism Severity (ß=.206, *p*<.05), General Adjustment Problems (ß=.396, *p*<001), having experienced unmet needs (ß=.140, *p*<05), and age (ß=-.210, *p*<.001) were associated with the total frequency of barriers endorsed after controlling for frequency of contact with the health services.

Patient-related barriers

Autism Severity (ß=.330, *p*<.001), General Adjustment Problems (ß=.492, *p*<.001), and younger age (ß=-.116, *p*<.05) were associated with the frequency of patient-related barriers endorsed, when controlling for the frequency of contact with the health services variables (Table 5).

HCP-related barriers

General Adjustment Problems (ß=.324, *p*<.01), age (ß=-.142, *p*<.05), and having experienced unmet needs (ß=.174, *p*<.05) were associated with the frequency of barriers endorsed at the HCP level after controlling for the health services contact variables (Table 5).

Healthcare system-related barriers

Anxiety (ß=.218, *p*<.05), having experienced unmet needs (ß=.184, *p*<.01) and younger age (ß=-.205, *p*<.01) were associated with the frequency of barriers endorsed at the healthcare system level, after controlling for frequency of contact with the health services (Table 5).

Barriers related to managing care

After controlling for the frequency of contact with the health services, General Adjustment Problems (ß=.239, *p*<.05), and younger age (ß=-.179, *p*<.05), were associated with the frequency of barriers endorsed related to managing care (Table 5).

Predictors of the total severity of barriers endorsed

A total of 121 participants were included in this analysis. General Adjustment Problems (ß=.471, *p*<.001) and younger age (ß=-.192, p<.01) were associated with the severity of barriers experienced (Table 5).

Table 5. Summary of regression analyses

	Variable	**Beta**	**SE**	***F change***	***B***	***R*²**	***A R*²**
Predictors of total frequency of barriers							
1.	GP visits	.945	.452	5.789**	.159*	.087	.72
	Hospital appointments	.536	.358		.108		
	ED visits	3.022	1.507		.151		
2.	Autism severity	.195	.069	14.810***	.206*	.483	.447
	Anxiety	1.439	3.921		.032		
	Depression	1.615	3.514		.039		
	OCD	-2.670	2.904		-.066		
	General adjustment problems	17.434	4.409		.396***		
	Unmet needs	7.030	2.903		.140*		
	Health status	-4.179	4.728		-.054		
	Age	-.586	.157		-.210***		
	Gender	-1.794	3.549		-.029		
Predictors of frequency of patient-level barriers							
1.	GP visits	.102	.169	2.385	.047	.038	.022
	Hospital appointments	.080	.134		.044		
	ED visits	1.161	.562		.160		
2.	Autism severity	.113	.026	14.177***	.330***	.445	.406
	Anxiety	-.697	1.476		-.043		
	Depression	-.189	1.323		-.013		
	OCD	-1.949	1.093		-.132		
	General adjustment problems	7.856	1.524		.492***		
	Unmet needs	-.540	1.093		-.030		
	Health status	1.704	1.780		.061		
	Age	-.118	.059		-.116		
	Gender	-.397	1.336		-.018		
Predictors of frequency of HCP-level barriers							
1.	GP visits	.175	.117	3.013*	.116	.047	.031
	Hospital appointments	.141	.093		.112		
	ED visits	.424	.389		.084		
2.	Autism severity	.032	.021	5.900***	.132	.270	.220
	Anxiety	-.430	1.179		-.038		
	Depression	.438	1.056		.042		
	OCD	-1.032	.873		-.100		
	General adjustment problems	3.602	1.217		.324**		
	Unmet needs	2.208	.873		.174*		
	Health status	-1.778	1.421		-.091		
	Age	-.100	.047		-.142*		
	Gender	-.923	1.067		-.059		
Predictors of frequency of healthcare system-level barriers							
1.	GP visits	.479	.182	5.507**	.201**	.083	.068
	Hospital appointments	.201	.144		.101		
	ED visits	.775	.606		.097		
2.	Autism severity	.032	.031	8.366***	.083	.360	.316
	Anxiety	3.898	1.751		.218*		
	Depression	-.286	1.569		-.017		
	OCD	-.003	1.297		.000		

	General adjustment problems	3.444	1.808		.195		
	Unmet needs	3.701	1.296		.184**		
	Health status	-2.711	2.111		-.088		
	Age	-.229	.070		-.205**		
	Gender	.113	1.585		.005		
Predictors of frequency of barriers related to management of healthcare							
1.	GP visits	.121	.070	3.419*	.134	.053	.038
	Hospital appointments	.074	.055		.099		
	ED visits	.290	.233		.096		
2.	Autism severity	.015	.013	4.370***	.108	.228	.174
	Anxiety	-1.198	.726		-.177		
	Depression	.888	.651		.142		
	OCD	.311	.538		.050		
	General adjustment problems	1.596	.750		.239*		
	Unmet needs	.381	.538		.050		
	Health status	-1.260	.876		-.108		
	Age	-.076	.029		-.179*		
	Gender	-.417	.657		-.045		
Predictors of overall severity of barriers							
1.	GP visits	.388	.326		.094		
	Hospital appointments	.254	.210		.088		
	ED visits	-.439	1.285	9.137***	-.026	.504	.449
	Autism severity	.083	.057		.136		
	Anxiety	1.809	3.166		.062		
	Depression	.527	2.784		.020		
	OCD	-1.318	2.493		-.048		
	General adjustment problems	13.477	3.350		.471***		
	Unmet needs	3.281	2.236		.103		
	Health status	-3.170	3.991		-.062		
	Age	-.319	.177		-.192**		
	Gender	.205	2.751		.005		

GP=general practice; ED=emergency department; OCD=Obsessive Compulsive Disorder

Discussion

Research suggests that individuals on the autism spectrum experience difficulties accessing the healthcare they need (9). This paper aimed to develop a valid and reliable caregiver-report Barriers to Healthcare tool, and to use this tool to examine barriers as perceived by caregivers of autistic individuals, as well as potential contributing factors.

A recent taxonomy of barriers to healthcare for autistic individuals indicated that although a number of caregiver-report tools exist, they fail to capture the barriers which may exist at the levels of the patient, HCP, and system (28). Therefore, the current tool aimed to fill this gap and address barriers at each level. It is important to note that there was some convergence with items from previously developed tools (e.g., 34, 51, 52). This is unsurprising as there are a finite number of ways to ask about a particular barrier, as evinced by the convergence observed across tools included in the previous systematic review (28); for example, “I don’t have insurance coverage” (34); “Insurance benefits never/always cover child’s needs” (53); “Difficulties related to insurance coverage” (54); “no insurance” (55). Since these studies, among others, informed the taxonomy upon which the tool was based, some level of convergence is to be expected. Fifteen of the original items were removed as part of the development of the tool, a common practice in survey tool development (50). The EFA indicated that the latent constructs underlying our tool did indeed corresponded to patient, HCP, and systems-related barriers. An additional factor also emerged, which does not fall easily onto any one of the three levels: barriers related to managing healthcare (e.g., finding it difficult to follow up on care). It is possible that this occurred as the tool was developed based on barriers identified/endorsed by caregivers, autistic individuals, and HCPs. For autistic individuals, difficulty on following up on care may be related to patient-related factors such as issues with executive function (34). For caregivers, this may reflect systemic issues (e.g., being unable to get referrals; 55), or HCP-related barriers (e.g., poor communication between the HCP and caregiver; 56). Future research could investigate this more thoroughly.

The second aim of this paper was to examine the barriers that were endorsed by participants. The findings that barriers occur at the patient, HCP and systems levels echo previous research (32, 33, 57). Difficulty reporting/identifying pain/symptoms was the barrier which was most often endorsed and echoes previous qualitative studies with autistic individuals, caregivers, and HCPs (57-59). Difficulty communicating/interpreting symptoms is a serious issue as misinterpretation can lead to diagnostic overshadowing, misdiagnosis, or delayed or non-receipt of care (57, 60, 61). To facilitate effective communication and interpretation of pain/symptoms, it is likely that a variety of interactive methods and observations are required (62). Choosing appropriate methods will depend on the autistic individual's specific needs and preferences but may include the use of electronic devices or rating scales such as the Wong-Baker FACES scale (62, 63).

More than half of caregivers reported a lack of HCP knowledge and training as a frequent barrier with over a third of caregivers identifying this as a severe issue. A lack of provider knowledge regarding autism has been widely reported by HCPs, caregivers and autistic individuals indicating the need for improved autism training for HCPs (64-66). Autistic standardised patients (SPs) are a novel method of incorporating autism training into the medical curriculum and encouraging results have been observed with nursing students when combined with classroom instruction (67, 68). Research should continue to investigate the conditions under which such simulations are most effective and assess their feasibility with other healthcare professionals.

The third aim of the paper was to examine the variables potentially associated with the frequency and severity of the endorsed barriers. Two of the key predictors were General Adjustment Problems and having experienced unmet needs in the past 12 months. General Adjustment Problems (e.g., aggression towards self/others) can hinder the delivery of care (52, 69). Such behaviours can be a response to feeling fearful or overwhelmed during a healthcare encounter but advanced preparation, use of rewards or reinforcement, and distraction or structured activities have been identified as helpful strategies for reducing the occurrence of such behavioural challenges in healthcare contexts (70, 71). Efforts should, therefore, be made to examine which strategies might work best for different individuals in preparation for a healthcare visit.

Similar to the current study, unmet needs have previously been linked to HCP-related barriers such as being unable to find HCPs who can adequately accommodate autistic patients (55). Providing training to HCPs which encompasses identifying and implementing accommodations could, therefore, reduce the occurrence of unmet needs. It is important to not only focus on HCP training, however. Altering the delivery of care may also reduce unmet needs. A growing body of research has indicated that access to a medical home (a team-based model of primary care which offers comprehensive and continuous, patient/family-centred care; 72) can reduce unmet needs and improve satisfaction with care (73). Since 2012, the Primary Care Centre (PCC) model has been developing in Ireland which is similar to the medical home model in the United States. PCCs aim to provide multidisciplinary, first-point of care which includes physician care, occupational therapy, speech and language therapy, and psychological services in one setting (74). The current tool could be used as a pre/post assessment to evaluate whether PCCs can reduce unmet needs and barriers to care for autistic individuals and their families in Ireland.

Limitations

There are a number of limitations to this study which need to be considered. First, despite sampling efforts, and although caregivers of autistic adults did participate, this group remained underrepresented and so the results of the study are likely more reflective of autistic children. Much of our recruitment was done through schools and online support groups which may be more likely to have younger parents as members. Future research could use the tool to compare barriers experienced by caregivers of autistic adults and children to assess whether separate tools would be more beneficial. Second, our sample of autistic individuals was predominantly male, but this is consistent with autism prevalence rates (75), future research may want to use to tool to compare barriers experienced by different genders. Third, autism diagnosis was caregiver-reported however, the use of the SRS-2 supported this, as over 90% of the sample scored above the cut-off score for indication of autism diagnosis (76) and those who did not, fell just marginally below it. Fourth, due to the correlational nature of regression analyses, only associations can be determined between the predictor and criterion variables without causal inference. Fifth, although the tool was informed by previous research with autistic participants, their supporters, and HCPs, it would have benefitted from direct community involvement. Unfortunately, this was not possible in the current project due to time and resource constraints. The authors support the need to engage in co-creation of tools and interventions with the autism community to ensure that research priorities are aligned, appropriate and acceptable (77) and recommend that future research conduct validation work on this tool with autistic self-advocates and their supporters. Sixth, although health insurance was listed as a barrier in the tool, it is unknown how many participants in the current sample had private health insurance. In two-tiered healthcare systems, such as in Ireland, private health insurance can be linked to higher socio-economic status, which can be linked to improved access to care (78, 79).

Future research may wish to use the current tool to compare barriers experienced by families who have private health insurance and those who do not. Seventh, no comparison group was included in the study; future research may want to use the current tool to compare barriers experienced by parents of autistic and non-autistic individuals. Finally, a high percentage of data were missing from the Severity of Barriers scale which was likely an unfortunate result of the presentation of the scale in the questionnaire. As a result of the missing data, the sample size was small for this particular scale so analysis of this data should be considered with caution.

Future research

It is important to acknowledge that this paper only describes the initial development of the tool and so further validation research is required to examine: reproducibility (does the same factor structure result from the analysis of another sample of responses?); responsiveness (is the tool sensitive to changes?): and interpretability (can qualitative meaning be assigned to the quantitative scores? (35). Future research could also further refine the tool and assess whether adaptations are required to suit different sub-groups (e.g., individuals with intellectual disabilities). Cognitive interviewing with autistic individuals and their caregivers may be beneficial for this.

We recommend that this tool be used to assess the barriers which might exist in healthcare facilities. Previous work has highlighted that although HCPs are aware that accommodations are required, they often struggle with knowing what accommodations are needed and how to implement them (80). HCPs could ask caregivers of their autistic patients to complete the current tool so that they can identify what barriers exist for that patient in their specific healthcare setting. This may facilitate conversations between the HCP, the caregiver and the autistic person around what accommodations might be helpful and how these might be implemented; for example, allowing the patient to wait outside until their consultation if they are unable to tolerate the waiting area. Future research might also consider using the tool to collect data across different services to identify service-specific barriers. This knowledge could help raise HCPs' awareness of barriers they may be more likely to encounter in their own contexts and help them identify the required accommodations. This could also help determine whether tailored tools or subscales for different settings and medical professions would be beneficial.

The current tool could also be used to compare barriers experienced by different subgroups of the autism community. Autism severity was significantly associated with the barriers endorsed by caregivers in the current study which suggests a need to examine how different subgroups (e.g., those with co-occurring psychopathology, different intellectual abilities, different abilities in daily living skills) within the autism community experience barriers to healthcare. Knowing whether some barriers and their associated accommodations are more common for different subgroups could help HCPs better prepare for visits with different patients. More research representing autistic adults who may not be able to self-report is also needed. This group is under-represented in research in general (81). In the current study, autistic adults accounted for just 16.5% of the sample, despite recruitment efforts and it is unknown how many were capable of self-reporting. Consideration of ways to better engage caregivers of autistic adults who can and cannot self-report is recommended.

The current Covid-19 pandemic has exacerbated the inequities faced by autistic individuals, compounding the need to highlight and overcome barriers to care (82, 83, 84). The increased social isolation has had disproportionate adverse effects on the mental health of autistic individuals and although many services have moved online, many people do not have access to, or the ability to use, the internet, so are not getting needed support (84). Many autistic people have underlying medical conditions, placing them at a higher risk of complications if they contract the virus; yet those living in residential care, or as inpatients in mental healthcare facilities, may be more at risk of becoming sick due to living in close quarters with others (83). Those who do become sick may face additional challenges. Communication issues, for example, may cause particular difficulties for an autistic person who is hospitalised if they cannot be accompanied by a supporter under coronavirus restrictions (83). On the other hand, the pandemic has also demonstrated how accommodations to services are possible. Changes in service delivery, such as the move to online/telephone healthcare consultations or waiting in the car outside a healthcare facility rather than in the waiting area, are examples of accommodations that the autism community have long been asking for, which have become the norm as a result of the pandemic (84). Future research should monitor the effects of such changes on the accessibility of healthcare for autistic individuals and examine how these changes may be maintained in the long term for those who need them.

Conclusions

Caregivers of autistic individuals indicate that a range of barriers are experienced by people on the autism spectrum when attempting to access healthcare, and that these barriers occur at the level of the patient, healthcare provider and system. As a result, the autistic community experiences disparities in healthcare and unmet healthcare needs. In order to improve access, initiatives which aim to make healthcare more inclusive for individuals on the autism spectrum, such as improved autism training for healthcare professionals, are required. It is hoped this tool will help to identify areas most in need of attention and guide intervention development and evaluation.

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Chapter 4: Study 3

Development and Preliminary Evaluation of a Novel Physician-Report Tool for Assessing Barriers to Providing Care to Autistic Patients

Development and preliminary evaluation of a novel physician-report tool for assessing barriers to providing care to autistic patients

Declaration

Where this fits in with the thesis

Considering the findings of Chapter 3, it is apparent that caregivers identify many healthcare provider (HCP)-related barriers. However, Chapter 2 demonstrated that no HCP-report tool exists which could assess barriers to care at the levels of the patient, HCP, and system. This is despite widespread agreement that HCP knowledge and self-efficacy with regards to caring for autistic individuals is low (1,2,3). Thus, valid and reliable tools which could facilitate a thorough assessment of barriers is required. The purpose of Study 3 was, thus, to develop a physician-report tool to address the research question: *what are the barriers to providing care to people on the autism spectrum experienced by physicians in Ireland?* This tool may be helpful for identifying barriers which occur in various healthcare settings and for identifying supports required by physicians to enable them to provide quality healthcare to their autistic patients.

Peer-reviewed publication

This study is currently under review in a peer-reviewed journal.

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Author contributions

CW, POC and SL proposed and designed the study. CW, RG, CC, MC, LO'L, EW & DB contributed significantly to data acquisition and interpretation . CW drafted the initial manuscript. analysed the data and drafted the manuscript. CW, SL, POC, CW, RG, CC, MC, LO'L, EW & DB contributed to the final manuscript. All authors have read, commented on, and approved the final manuscript.

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Abstract

Background. Individuals on the autism spectrum face significant disparities in health and physicians often report difficulties in providing care to autistic patients. In order to improve the quality of care autistic individuals receive, it is important to identify the barriers that physicians experience in providing care so that these may be addressed. This paper reports the initial development and preliminary evaluation of a physician-report ‘Barriers to Providing Healthcare’ measurement tool.

Method. An established taxonomy of healthcare barriers for autistic individuals informed the initial draft of a 22-item measurement tool. This measurement tool was distributed to physicians working in various healthcare specialties and settings. Exploratory factor analysis (EFA) was conducted to determine the construct validity of the tool; discriminant validity between, and internal consistency of, the resultant factors were assessed. Multiple regressions were used to explore variables potentially associated with barriers endorsed by physicians.

Results. A total of 203 physicians were included in the analyses. The EFA resulted in a 17-item tool with three distinct factors which explained 37.6% of the variance: 1) Patient-related barriers (Cronbach’s α=0.83; e.g., the patient’s reactivity to the healthcare environment); 2) Healthcare provider (HCP)/family-related barriers (Cronbach’s α=0.81; e.g., a lack of providers willing to work with autistic patients); and 3) System-related barriers (Cronbach’s α=0.84; e.g., there is a lack of support for patients and families). Discriminant validity between the factors was adequate ($r<0.8$). The barriers that were most frequently endorsed as occurring ‘often’ or ‘very often’ included a lack of support for patients and families (endorsed by 79.9% of physicians); communication difficulties (73.4%); and a lack of coordination between services (69.9%). The regression analyses identified no significant associated variables.

Conclusion. A preliminary version of a novel physician-report tool to assess barriers to providing care to autistic patients has been developed although further validation work is required. The use of this tool will help physicians to identify issues specific to different medical specialities and healthcare settings. This information may help identify the supports physicians require to recognise and implement the required accommodations. Future research which elucidates barriers to healthcare provision for autistic patients is required to support systemic change in healthcare so as to improve care experiences and health outcomes for people on the autism spectrum.

Keywords: Autism; Physicians; Healthcare Access; Health Equity; Reasonable Adjustments; Health.

Conflict of interest

The authors state they have no competing interests.

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Introduction

Substantial inequities in health outcomes and healthcare access exist for people with disabilities (1, 2). These inequities are not attributable to an individual's disability, however, but rather reflect a disparity in the accessibility of healthcare, whereby people with disabilities have more difficulty accessing the healthcare they need than others (3). Autism Spectrum Disorder (hereafter 'autism') is one specific condition in which significant health disparities exist (4, 5).

Autism is a neurodevelopmental condition characterised by persistent difficulties or differences in social interactions and communication alongside repetitive and/or restrictive patterns of behaviour (6). Currently, prevalence is estimated at one in 54 children, and one in 45 adults in the USA (7, 8). Global prevalence is currently estimated at approximately 1 in 160 persons and is rising (9). Autistic[3] individuals experience a high prevalence of medical and psychiatric co-occurring conditions, which can make care complex (11). This may be one reason that autistic individuals present to both emergency and non-emergency healthcare services more often (12), are admitted to hospital more often (13), and have longer hospital stays than their neurotypical peers (14). Yet, despite a higher rate of healthcare utilisation, autistic individuals tend to experience more unmet healthcare needs (15, 16), poorer health outcomes (11, 17), poorer healthcare related quality of life (18, 19) and higher

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[3] Use of identity-first language: many autistic individuals have indicated a preference for the use of identity-first language (i.e., autistic person) as opposed to person first language (i.e., person with autism; 10). Accordingly, this article will use identity first language.

mortality rates than others (20, 21). These data suggest that a disparity in access to quality healthcare exists for autistic individuals (22).

A number of recent systematic reviews have identified a variety of complex barriers which exist for autistic individuals when accessing and receiving healthcare which are likely contributing to this disparity (23-25). These barriers may be understood to exist across the healthcare system: at the level of the patient (e.g., barriers associated with autism-related characteristics such as communication difficulties), healthcare provider (e.g., insufficient autism knowledge), and the system/organisation (e.g., a lack of resources) (23).

Considering the high prevalence of co-occurring conditions, the poorer healthcare experiences of autistic individuals, and the multitude of barriers which are known to exist, there is a recognised and pressing need to improve healthcare services for this population (15, 26). Indeed, autistic self-advocates have highlighted improving health services and physical health outcomes as a research priority (27).

Ensuring accessible and equitable healthcare for autistic individuals, as is legally mandated in the UK (28, 29), requires that physicians are equipped with the tools, knowledge, and resources they need to care appropriately for this patient population (30). In order to ensure that this is the case, the specific difficulties physicians face when caring for their autistic patients first need to be understood. Although measures exist to facilitate exploration of healthcare-associated barriers among autistic adults (26), or the caregivers of autistic individuals (31), there is a lack of established methods for engaging staff about their experiences and perceptions. A number of previous studies have used HCP-reported measurement tools to assess barriers to providing healthcare to autistic individuals, however, they are typically limited in the array of barriers that they assess (e.g.,32-34). HCP-reported tools are important because they can provide information which may be missed by relying on the patient or caregiver perspective alone. Further, many interventions developed to reduce barriers to healthcare for autistic individuals will likely take place in healthcare settings and so, the HCP perspective is needed to ensure that such interventions are feasible from a physician perspective and are targeting issues that are important to both physicians and autistic individuals.

Accordingly, the aims of this study were to: 1) develop a physician-report tool to assess a more comprehensive array of barriers which may occur across the healthcare system and report on some preliminary assessments of validity and reliability (Phase one of the current study); 2) to examine the barriers endorsed by physicians, and identify potential factors associated with these barriers (Phase two of the study).

Methods

Context

In Ireland, although there are a very small number of private sector mental health services for autistic individuals, there are currently no speciality clinics within physical healthcare for autistic individuals. Instead, autistic individuals attend physical healthcare services, and are cared for, in the same manner as the rest of the population.

Design

This study used a cross-sectional survey design.

Participants

Eligible participants were physicians working in primary, secondary, psychiatric, or tertiary care services across the Republic of Ireland, who had some experience of caring for autistic individuals. Guidance on the required sample size to conduct an EFA varies (35-37). A common rule of thumb denotes a 10:1 ratio of participants to items, though in general, larger sample sizes are recommended to produce stable factor structure (38).

Therefore, the authors aimed to satisfy this rule of thumb at the minimum but to recruit as large a sample as possible to ensure sufficient power.

Recruitment

A variety of non-probability sampling methods and recruitment strategies were employed in an effort to recruit as wide a sample as possible. Convenience sampling and voluntary response sampling included: 1) circulating emails among staff within one medical school and one hospital group comprising of six hospitals; 2) placing advertisements in local and national newspapers, on local radio, and on social media; and 3) snowballing methods whereby participants were asked to share information about the study with others in their organisation(s) and/or social network. Anyone interested in the study contacted the researcher to request a survey pack or a link to an electronic version of the questionnaire.

Data Collection

Participants who completed paper forms were provided with pre-paid stamped return envelopes to return their surveys to the researchers. As an incentive, participants were offered the opportunity to enter a prize draw to win one of four gift vouchers to the value of €50 each.

Ethical considerations

Ethical approval for this study was granted by the NUIG Research Ethics Committee (ref: REC 18-Jun-17). Informed written consent was obtained for each participant. For online surveys, participants clicked ‘I agree’ on an online consent form before being brought to the survey page.

Phase 1. Tool development and evaluation

Literature review

A systematic literature review of 31 studies, was conducted to identify barriers to healthcare for autistic individuals and to develop a taxonomy of those barriers (23). Included studies used quantitative, qualitative, and mixed methods, and participants included autistic individuals, caregivers, and HCPs. For quantitative studies, all questionnaire items which had been endorsed as barriers were extracted, while in qualitative studies, direct quotes and author-reported themes which described barriers were extracted. A thematic analysis approach was undertaken, initially guided by the domains outlined in Raymaker et al.'s (26) Barriers to Healthcare Tool. These domains included barriers related to: emotions, executive function, healthcare navigation, provider attitudes, patient-provider communication, sensory sensitivities, socio-economic issues, support, and waiting. A total of 320 individual barriers were identified across the studies. Not all barriers could be organised within the Raymaker et al. (26) domains and so new themes and subthemes were developed and organised into a taxonomy which consisted of three over-arching themes: 1) Patient-related barriers (autism-related characteristics; other patient-related barriers); 2) HCP-related barriers; 3) system-related barriers). Each theme had between two and eight subthemes (Table 1). Full details of this process are described elsewhere (23). These themes and subthemes informed the item construction for the current tool.

Table 1. Themes and subthemes within the taxonomy which guided item development

Theme	Subtheme
Patient-related barriers: Barriers associated with autism-related characteristics	Communication/social difficulties
	Issues with waiting
	Issues with executive function
	Sensory issues
	Anxiety/other emotion
	Need for consistency
	Behavioural issues
Other patient-related barriers	Complexity of family involvement
	Scepticism towards conventional medicine
HCP-related barriers	Lack of autism knowledge/skill
	HCP inflexibility
	Stigma/negative perceptions
	Difficulties interpreting behaviour/symptoms
	Ignoring patient/caregiver concerns/expertise
	Poor HCP communication/failure to adapt language
System-related barriers	Lack of support for patients/caregivers
	Lack of support for HCPs
	Time/resource constraints
	Lack of continuity/collaboration between HCPs/services
	Location issues
	Financial/insurance issues
	Lack of qualified personnel
	Inflexible HC system

HCP=Healthcare provider; HC=Healthcare

Item construction

Best practice in questionnaire design guided the development of the items for instrument development (39). An iterative method was used to construct the items of the questionnaire. This involved two consensus building meetings between three members of the research team (CW, SL, POC). Specific items from existing questionnaires focused on barriers to healthcare for autistic patients were deliberately not reviewed as part of the item construction process as the team focused on the themes and subthemes of the taxonomy which had resulted from the prior systematic review (23). In the first meeting, questionnaire items (*n*=45) were constructed. The items were then reviewed, refined, and condensed into 22 items, each representing a different barrier linked to a subtheme. This process involved the research team discussing the items in detail and working together to identify items which were measuring the same subtheme. During a subsequent meeting, this process of reviewing the items was repeated to ensure all previous decisions were confirmed and to make any final refinements. This initial reduction in the number of items was conducted in order to avoid repetition among items and to reduce respondent burden. This is in line with best practice which recommends that measurement tools should be ‘usable’ (i.e., short, readable, and easy to complete) in order to avoid the potential for errors or non-response (40). Efforts were made to reflect barriers which might occur at the level of the patient, HCP, and system**.**

Frequency and Severity of barriers

For each item within this tool, respondents were asked to indicate the frequency with which each barrier had occurred in the past 12 months, rated on a Likert scale of 0 (never) to 4 (very often). Although it is generally recommended (41), a more concrete frequency scale (e.g., hourly, weekly, monthly etc.) was not utilised as it was expected that participants would have varying levels of contact with autistic patients. This made it difficult to frame frequency of contact around specific time periods. The options of 'hourly or weekly' for example, would not be applicable to a physician who only sees autistic patients occasionally. However, if a particular barrier was encountered every time the physician met an autistic patient, even if they only occasionally meet autistic patients, this might be considered a frequent barrier. Therefore, it was not deemed appropriate to use objective time intervals as these would only work if all participants were seeing autistic patients with roughly the same frequency. A recall period of 12 months was chosen as, although a shorter recall period (e.g., six months) may improve recall performance in some cases, such short recall periods may not capture infrequent events or behaviours (42-44). As the respondents were anticipated to have fairly infrequent interactions with autistic patients, a 12 month recall period was chosen as the most appropriate recall period for this study.

The level of perceived severity (i.e., how much of a problem the barrier presented for the respondent) each barrier posed was rated on a Likert scale of 1 (slight), 2 (moderate), or 3 (severe). The response options were adapted from the Behaviour Problems Inventory (45) and the way in which the responses were presented in the tool was modelled on the Behaviour Problems Inventory. This presentation was chosen as it allowed assessment of both frequency and severity in tandem, rather than repeating the survey items twice (i.e., once for frequency and again for severity). It was hoped this would place the least burden on the respondent.

Statistical analyses

All statistical analyses were conducted in IBM SPSS (version 21). Significance levels for all analyses were set as $p<.05$.

Factor analysis

Factor analysis refers to a set of statistical procedures that can be used to identify the underlying constructs or domains that exist in a tool that is being developed (39, 43). Factor analysis can take two forms: Exploratory Factor Analysis (EFA) and Confirmatory Factor Analysis (CFA). An EFA was chosen for this analysis rather than a CFA for a variety of reasons. First, it is both common and recommended that, on initial development of a new tool, an EFA should be conducted as a first step in assessing construct validity of a measure, even when existing literature and *a priori* hypotheses regarding factor structure to guide tool development exists (39, 46-48). Second, EFA is used to identify latent constructs when there is insufficient evidence to make strong assumptions about the relationships among the items, how many common factors exist or what specific variables these common factors are likely to influence (46, 49, 50), as was the case in the current study. CFA, on the other hand, is conducted when a substantial theoretical base already exists, or when the relationship between items has already been tested and the factors and related items are known (51, 52). CFA is typically used after an EFA, with a new data set, to assess the goodness of fit of a model when there is a strong model assumption (53). Third, our aim was not to test the taxonomy, but to continue to refine the theory surrounding barriers to care. Thus, EFA was considered the more appropriate analysis to undertake in the current study due to the exploratory nature of the study, a lack of a sufficiently strong theoretical assumption of the model structure and the relationship between the items, and our aim of refining theory rather testing it (46, 49).

Initial data screening

Little’s test for Missing Completely at Random, and Missing Data Analysis as applied in IBM SPSS was used to assess the missing data.

Construct validity

Exploratory factor analysis (EFA), a reduction technique that enables the determination of the common latent variables that underlie the various items in a scale (54) was used to determine the construct validity of the Barriers to Providing Healthcare tool developed by the researchers. The EFA was conducted in accordance with best practice (38), and proceeded through the following steps:

Step 1: Adequacy of the correlation matrix.

Suitability of the data to an EFA was assessed by considering the sample size, factorability of the constructs (correlation matrix), examination of the Kaiser-Meyer-Olkin (KMO) Measure of Sampling Adequacy (MSA) and Bartlett's Test for Sphericity.

Step 2. Factor extraction, retention, and interpretation.

Principal Axis Factoring was chosen as the factor extraction method as this method has an explicit focus on latent factors, whereas principal component factors, another common method, is computed without regard to any underlying structure caused by latent variables (38). Factor extraction was determined by considering Kaiser's criteria (Eigenvalue >1), the scree plot, and a parallel analysis (PA) which was conducted via an online PA engine (55). Oblique (Promax) rotation was used as the data cannot be assumed to be completely independent of each other and this is considered most accurate for research involving human participants (38, 48). Through an iterative process, items were removed if they loaded onto more than one factor with a value ≥ 0.4 or had weak loading values of <0.4 (56, 57). The pattern matrix guided interpretation and naming of the factors by the research team (56, 58).

Step 3. Discriminant validity and internal consistency of the factors
Discriminant validity between the generated factors was assessed by examining the factor correlation matrix, with values <0.8 indicative of adequate discriminant validity. Internal consistency of each of the generated factors was assessed using Cronbach's alpha, with values >0.7 indicative of good internal consistency (59).

Phase 2. Assessment of barriers

Measurement tool

The survey instrument administered to participants consisted of three sections: 1) perceived barriers to providing care to autistic patients (i.e., the novel tool developed in Phase One); 2) physician knowledge of autism; and 3) demographics.

Frequency/severity of barriers

The measurement tool described in Phase one was administered. The tool contained 18 items which corresponded to individual barriers. Participants were asked to rate the perceived frequency and severity of each barrier presented. Subscale scores were calculated by summing the items in each subscale with higher score indicating more problems with the barriers.

Knowledge of autism

To assess participating physicians' knowledge of autism, a 22-item Knowledge of Autism Scale, which uses a 'true/false' response option, was used (60, 61). This scale assesses the participant's knowledge of early signs of autism, descriptive characteristics, and commonly co-occurring behaviours. To score the scale, eight items are reverse scored and then all items are summed to obtain a total scale score. A score of 1 is attributed to true answers and a score of 0 to false answers. This scale has previously demonstrated moderate internal consistency (Cronbach's α=0.54; 61). The internal consistency of the Knowledge of Autism scale in the current study was assessed by calculating Cronbach's alpha.

Demographics

Physicians were asked to provide information on their gender, years of clinical practice, medical specialty, prior training in relation to autism, and the approximate number of autistic patients they treat per annum (Table 2).

Statistical analyses

All statistical analyses were conducted in IBM SPSS (version 21). Significance levels for all analyses were set as $p<.05$.

Missing Data analysis

Littles Test for Missing Completely at Random was used to assess the missing data. Where data were missing, simple mean imputation was used to replace the missing values (62) which allowed for the production of subscale scores needed for analysis.

Factors associated with barriers endorsed

In order to assess variables potentially associated with barriers, three hierarchical multiple regressions were conducted. Preliminary analyses were performed to ensure no violations of the assumptions of normality, multicollinearity, and homoscedasticity. The regressions assessed whether the frequencies of barriers were associated with the following variables: 1) medical specialty; 2) years since graduation from medical school; 3) attendance at autism training; and 4) autism knowledge. The number of autistic patients seen per year was controlled for within the regressions as it might be expected that physicians who see a higher number of autistic patients may report a higher frequency of barriers. The same method was used to complete all three regressions with just the criterion variable (i.e., patient-related barriers subscale; HCP/family-related barriers subscale; system-related barriers subscale) changed in each case.

Results

Response rate

As a variety of recruitment methods were used (e.g., leaflets, social media), it is not possible to provide an entirely accurate response rate. However, a total of 400 paper surveys were distributed and 226 were returned– an estimated response rate of 55.6%.

Participants

A total of 23 participants did not provide any data for the Barriers to Providing Healthcare section and so were removed from all analyses, leaving a final sample of 203 physicians. The characteristics of these participants are presented in Table 2.

Table 2. Respondent characteristics

Respondent Characteristics	***N* (%)**		***N* (%)**
Gender		*Medical specialty of participants*	
Female	116 (57.1)	General practice	37 (18.2)
Male	83 (40.9)	Paediatrics	28 (13.8)
Prefer not to say	2 (1.0)	Psychiatry	27 (13.3)
Other	2 (1.0)	General internal medicine	20 (9.9)
Level of seniority		Surgery	10 (4.9)
Intern	34 (16.7)	Geriatrics	7 (3.5)
SHO	46 (22.7)	Neurology	6 (3.0)
Registrar	53 (26.1)	Emergency medicine	4 (2.0)
GP Trainee	3 (1.5)	Anaesthesia	4 (2.0)
GP	34 (16.7)		
Consultant	33 (16.3)		
Years since graduation			
<5	81 (39.9)		
5-10	54 (26.6)		
11-20	39 (19.2)		
21-30	11 (5.4)		
>30	18 (8.9)		
Autism training received			
Undergraduate education	95 (46.8)		
Postgraduate education	53 (26.1)		
Continued medical education	42 (20.7)		
Other	12 (5.9)		
Never	49(24.1)		
No. of autistic patients annually			
<10	142 (70.0)		
10-30	41 (20.2)		
31-60	17 (8.4)		
61-100	2 (1.0)		
>100	1 (.5)		

Note: Numbers under the autism training category do not add to 203 because participants could choose more than one option; Numbers under medical specialty do not equal 203 because not all respondents provided this information and interns are not included in this category; Levels of seniority are listed in ascending order.
GP=general practitioner; SHO=senior house officer

Phase 1. Tool development and evaluation

Initial data screening

Of the Frequency of Barriers scale, 8.4% of the data were missing. Little's test of missing completely at random indicated however, that the data were missing at random (χ^2=249.162, *df*=250, *p*=.503). No items were highly skewed or kurtosed (i.e., <-2/>2; 59, 62). Simple mean imputation was therefore used to replace the missing data (62). Inspection of the Severity scale indicated that a large amount of data were missing per item (*M*=18.45%; *SD*=4.07%; range 14%-30.5%) and this appeared to be an artefact of the way the scale had been presented. This was considered to compromise the data that resulted from this scale. Therefore, the EFA was run using the Frequencies of Barriers data only.

Construct validity

Assessment of the adequacy of the correlation matrix

On examination of the correlation matrix two pairs of items were highly correlated (>0.7). As a result, two items were deleted, one from each pair. The retained items made more theoretical sense based on barriers more commonly reported in the extant literature (e.g., a lack of coordination between services was retained instead of a lack of access to autism specialists). Subsequent examination of the correlation matrix suggested multicollinearity was unlikely to be an issue (58). Bartlett's test of sphericity indicated the correlation matrix was not an identity matrix (χ^2=213.836, *df*=190, *p*<.001). The Kaiser-Meyer-Olkin (KMO) Measure of Sampling Adequacy also indicated that the matrix was suitable for EFA (KMO=0.91). Examination of the MSAs along the principal diagonal of the anti-image correlation matrix indicated that all items were suitable for inclusion in the EFA as all had a value greater than 0.8 or 0.9, and all off-diagonal values were small (<0.2; 35).

Factor extraction, retention, and interpretation

Based on the Eigenvalue >1 rule, five factors were extracted, the Parallel Analysis extracted three factors and the scree plot extracted 2 factors. The Kaiser Eigenvalue >1 rule is not recommended as it has a tendency to over extract factors (35). Therefore, follow up analyses were run extracting: 1) two factors based on the Scree plot, and 2) three factors based on the Parallel Analysis. When situations arise in which the various procedures suggest different numbers of factors, or when the procedures produce somewhat ambiguous results, it is recommended that the researcher examine the subset of models produced to assess which solution produces the most readily interpretable and theoretically sensible pattern of results (46). After careful consideration of the two produced models, the three factor model produced by the Parallel Analysis made more theoretical sense as the items that clustered together were better interpreted as patient-related, HCP/family-related, and system-related. The two factor model is presented in Appendix 3.1. Correlations between the factors were observed in the factor matrix suggesting that an oblique rotation (Promax) be maintained (58). A number of iterations of this analysis were conducted to identify and remove redundant items and to ascertain the best model for the data. On the first iteration, one item loaded very similarly onto 2 factors and so was discarded from the analysis. On the second iteration, one item did not load onto any factor >0.4 and so was discarded from the analysis. On the third iteration, one item cross loaded on to two factors with a difference of <0.2 and so was discarded from analysis. On the next iteration all items had factor loadings >0.4 with no cross loadings evident, so no further iterations were conducted. This final model explained 44.3% of the variance. Table 3 presents the three extracted factors with the corresponding items, factor loadings and the amount of variance explained by each factor. The items which were removed during the analysis are presented in Appendix 3.2.

Determination of discriminant validity and internal consistency of the generated factors

The factor correlation matrix indicated adequate discriminant validity between the two factors as the values were all <0.8. As can be seen in Table 2, Cronbach's alphas indicated that all factors showed good internal consistency (all >0.8) as per conventional standards of interpretation (63).

Table 3. EFA of three factor solution based on parallel analysis

	Factor 1	**Factor 2**	**Factor 3**
Patient-related barriers. Cronbach's alpha= 0.83 Variance explained: 37.6%			
Challenging behaviours exhibited by patient	**.808**		-.186
The patient's reactivity to the healthcare environment	**.797**	-.172	
There are communication difficulties	**.733**		
Lengthy waiting room times for patients on the autism spectrum	**.550**		
The patient's use of outside providers	**.518**	.286	
Consultations are too short to accommodate patients on the autism spectrum	**.476**		.259
HCP/family-related barriers. Cronbach's alpha = 0.81; Variance explained: 5.9%			
There is a lack of clarity regarding GP remit/referral		**.717**	
There are financial disincentives due to the need for additional time with the patient.	.299	**.658**	-.223
The patients' family/caregivers are sceptical of conventional medicine (e.g., vaccines).		**.602**	
There is a lack of providers willing to work with patients on the autism spectrum		**.586**	.237
Family/caregiver involvement makes provision of healthcare to patients on the autism spectrum more complex		**.561**	.146
I prefer to avoid working with patients on the autism spectrum	-.158	**.554**	
System-related. Cronbach's alpha = 0.84; variance explained: 5.6%			
There is a lack of support for families and patients			**.814**
The physical environment in healthcare settings is unsuitable	.170	-.102	**.719**
Lack of own knowledge for working with patients on the autism spectrum	-.191	.164	**.614**
There is a lack of coordination between services	.238		**.584**
There are shortages of medical and non-medical services for autistic individuals	.272	.154	**.528**

GP=General practitioner

Phase 2. Assessment of barriers

Frequency scores

As can be seen in Table 4, the barriers most endorsed as occurring ‘often’ or ‘very often’ included: a lack of supports for patients and families (79.9%); communication difficulties (endorsed by 73.4% of respondents); and a lack of coordination between services (69.9% of respondents). More details on barriers endorsed are provided in Table 4.

For the novel tool, subscale scores were calculated by summing the responses for each subscale produced by the EFA (i.e., Patient-related barriers; HCP/family-related barriers; system-related barriers). On average, physicians scored highest on the patient-related barriers subscale (*M*=14.8, *SD*=5.0; range 0-24). This was followed by the than the system-related barriers subscale (*M*=13.8, *SD*=4.3 range:0-20). HCPs scored lowest on the HCP/family-related subscale (*M*=10; *SD*=5.5; range:0-24).

Severity scores

Due to a large amount of missing data on the severity scale, no analysis was conducted using these data.

Knowledge scores

Missing data analysis indicated that 10% of the data were missing from the knowledge scale. Little’s test of Missing Completely at Random indicated that the data were missing at random (χ^2=207.519, *df*=224, *p*=.778). Therefore, simple mean imputation was used to replace the missing values (62). Internal consistency was deemed moderate (Cronbach’s α=0.58). Physicians generally scored highly on autism knowledge (*M*=18.5, *SD*=2.2, range: 9-22).

Table 4. Number and percentage of respondents who endorsed each item

	Very often *N*(%)	**Often** *N*(%)	**Sometimes** *N*(%)	**Rarely** *N*(%)	**Never** *N*(%)
Patient-related barriers					
Challenging behaviours exhibited by the patient	17(8.4)	94(46.3)	62(30.5)	23(11.3)	7(3.4)
The patient's reactivity to the healthcare environment	28(13.8)	108(53.2)	42(20.7)	17(8.4)	8(3.9)
There are communication difficulties	57(28.1)	92(45.3)	31(15.3)	17(8.4)	6(3)
Lengthy waiting room times for patients on the autism spectrum	36(17.7)	67(33)	50(24.6)	23(11.3)	27(13.3)
The patient's use of outside providers	24(11.8)	67(33)	56(27.6)	18(8.9)	38(18.7)
Consultations are too short to accommodate patients on the autism spectrum	38(18.7)	68(33.5)	53(26.1)	21(10.3)	23(11.3)
HCP/Family-related barriers					
There is a lack of clarity regarding GP remit/referral.	26(12.8)	55(27.1)	50(24.6)	22(10.8)	50(24.6)
There are financial disincentives due to the need for additional time with the patient.	19(9.4)	42(20.7)	49(24.1)	28(13.8)	65(32)
The patients' family/caregivers are sceptical of conventional medicine (e.g., vaccines).	9(4.4)	29(14.3)	63(31)	59(29.1)	43(21.2)
There is a lack of providers willing to work with patients on the autism spectrum	21(10.3)	46(22.7)	51(25.1)	33(16.3)	52(25.6)
Family/caregiver involvement makes provision of healthcare to patients on the autism spectrum more complex	22(10.8)	39(19.2)	56(27.6)	50(24.6)	36(17.7)
System-related barriers					
There is a lack of support for patients/families	70(34.5)	82(40.4)	25(12.3)	15(7.4)	11(5.4)
The physical environment in healthcare settings is unsuitable	53(26.1)	87(42.9)	38(18.7)	14(6.9)	11(5.4)
Lack of own knowledge for working with patients on the autism spectrum	34(16.7)	96(47.3)	40(19.7)	23(11.3)	10(4.9)
There is a lack of coordination between services	65(32)	77(37.9)	29(14.3)	19.(9.4)	13(6.4)
There are shortages of medical and non-medical services for autistic individuals	45(22.2)	100(49.3)	29(14.3)	13(6.4)	16(7.9)

GP=general practitioner

Variables potentially associated with barriers

Assumptions of regression

Preliminary analyses indicated no violations of the assumptions of multicollinearity and homoscedasticity.

Variables associated with the frequency of patient-related barriers

The overall model, which included autism knowledge, medical specialty, years since graduation, and previous autism training, was not significant. Details of these findings are provided in Appendix 3.3.

Variables associated with the frequency of HCP/family-related barriers

The overall model, which included autism knowledge, medical specialty, years since graduation, and previous autism training, was also not significant (see Appendix 3.3).

Variables associated with the frequency of system-related barriers

The overall model, which included autism knowledge, medical specialty, years since graduation, and previous autism training, was also not significant (see Appendix 3.3).

Discussion

In order to reduce inequities in access to healthcare there is a need to make adaptions to health services and the delivery of care to accommodate autistic patients. In some countries, including the UK, this has become a legal requirement (28, 29). However, physicians and healthcare organisations struggle to identify how best to adapt services or support autistic patients (30, 64, 65). This paper reports the development of a novel physician-report Barriers to Providing Healthcare measurement tool that may be used to identify priority areas for change and which can support quality improvement activities. The data collected provide an important insight into physicians' experiences of barriers to providing care to autistic patients.

Phase 1. Tool development and evaluation

The EFA resulted in a 17-item tool for the assessment of barriers physicians experience when providing healthcare to their autistic patients. The tool consists of three subscales: patient-related factors (e.g., communication difficulties); HCP/family-related factors (e.g., HCP prefers to avoid working with autistic patients), and system-related factors (e.g., there is a lack of coordination between services) that demonstrate good internal consistency and construct validity. This tool is largely congruent with current understandings of barriers to healthcare access experienced by autistic patients and their caregivers (26, 66), and may be considered to supplement existing tools for caregivers (31) and autistic adults (26). It must be noted that there is some convergence between items in the current tool and those of previously reported tools which assess barriers to healthcare for autistic patients (e.g., 13, 33, 61, 67). This is to be expected since the items were based on a taxonomy that was informed by the existing literature and there is only a finite number of ways in which to describe a given barrier.

The current data align relatively well with the systematic review derived taxonomy (23) used to guide the tool development. The systematic review found three distinct themes: 1) patient-related barriers (split into a) barriers associated with autism-related characteristics, and b) other patient-related barriers); 2) HCP-related barriers; and 3) system-related barriers). Although the EFA in the current study also produced three factors, some discrepancies emerged in how some items were organised within the factors resulting from the EFA. For example, lengthy waiting room times and consultations being too short loaded on to the patient-related factor when it might have been expected that they would load on to the HCP- or system-related factor. This suggests that physicians in the current sample perhaps interpret these factors as patient-related because, although they might exist for all patients, these barriers become a much more significant problem, and impact on care more greatly, when associated with autistic patients in comparison to non-autistic-patients. This added complexity specifically related to autistic patients has been commonly described by HCPs as a challenge in the literature (68). Further, a lack of autism knowledge among HCPs loaded onto the system-related factor, whereas in the taxonomy, this is an HCP-related factor. It is possible that HCPs interpret this as a system-related factor as they potentially feel that they do not have access to adequate training (61). A lack of clarity over GP remit/referral pathways, and financial disincentives both loaded onto the HCP/family-related factor when they might have been expected to load onto the system-related factor. It is unclear why this occurred, but the findings support previous research that has indicated that clinicians are sometimes less likely to see the system-related causes of problems (69). This suggests that more work is needed to help physicians identify and interpret system-related issues (69). Finally, two items relating to the involvement of caregivers (i.e., family/caregiver involvement makes care complex; family/caregivers are sceptical of conventional medicine) both loaded with HCP-related items to create the HCP/family-related factor. It is possible that HCPs perceive caregivers as a type of care provider because caregivers tend to be so highly involved in the provision of, and decision-making regarding, the care of autistic individuals across the lifespan. These discrepancies indicate the need for further validation work on both the current tool and the taxonomy. For example, another EFA could be conducted with a larger sample to see if the current factor structure remains stable. A CFA could also be conducted to test the factor structure.

Phase 2. Assessment of barriers

A recent taxonomy (23) elucidated how barriers to healthcare for autistic individuals may occur at various levels of the healthcare system. These findings are supported by the current study as physicians endorsed barriers at the level of the patient (e.g., communication issues), the HCP (e.g., HCPs unwilling to work with autistic patients), and the system (e.g., a lack of coordination between services). A lack of support for patients and families was most endorsed as occurring ‘often’ or ‘very often’ by HCPs in the current sample. This is consistent with existing literature. Warfield et al. (70), also reported that physicians in their study indicated that a lack of services and supports for autistic youth and adults was a challenge to providing care. Similar results were observed by Unigwe et al. (61) where physicians reported that their autistic patients were left unsupported due to a lack of joined up services which made referral difficult. In particular, GPs reported that there was a substantial lack of support for adults after receiving an autism diagnosis, and no supports for autism management or accessing therapy (61). Some HCPs have suggested that practices could compile a list of local supports that could be given to their patients as one means of addressing this barrier (70). However, adequate supports first need to be put in place. Thus, it is likely that system-level changes are required to support autistic individuals and their families, and such changes must also consider how to improve referral pathways to any such supports (61).

Communication difficulties or differences are a diagnostic criterion for autism and can certainly become barriers in medical contexts (6); for example, some individuals have difficulty expressing pain/symptoms (71) making interpretation difficult for the physician and/or caregiver (72, 73). Further, some individuals may have slower language processing speeds, making it difficult to keep up with the typical pace of a conversation (26) and may result in the patient missing important medical information or not having ample time to ask questions (74). Hospital passports, which allow the patient to communicate important information to the physician/healthcare facility (e.g., medications, allergies, sensory sensitives, or communication needs), have been shown to be helpful for facilitating communication between providers and patients with intellectual and developmental disabilities (75). Although autistic self-advocates have endorsed hospital passports, with the National Autistic Society UK (76) providing one version on their website, more empirical work is needed to examine the effectiveness of using hospital passports in improving communication between providers and their autistic patients.

A lack of coordination between healthcare services was the third most endorsed barrier. Due to the high prevalence of co-occurring conditions that many autistic individuals experience (11), their care can be complex and often requires the involvement of a variety of professionals, including mental health professionals, occupational therapists, physical therapists, speech and language therapists, social services, neurologists, and other clinical specialists (72). Several studies have highlighted issues with care coordination in the context of autism. For example, parents of autistic children were three times more likely to identify issues with care coordination between specialty doctors and other providers than parents of children with other types of special healthcare needs (77). Relatedly, physicians have noted difficulties such as identifying appropriate referral pathways as particularly problematic in relation to autistic patients (61, 72). Addressing such barriers will require system-level changes in how information is shared across healthcare services and providers. Electronic health records and flagging systems have shown promising results in improving the flow of information (78) between providers and may warrant further investigation in relation to autistic patients (78-80). The current tool could be used to assess in which contexts (e.g., primary care, neurology) care coordination is a more significant barrier and is therefore most in need of such interventions.

It was hoped that examining variables potentially associated with barriers (e.g., autism knowledge, years of clinical practice) would help to indicate where and for whom barriers are likely to occur so as to allow improvement efforts and resources to be most clearly directed. However, no clear associated variables emerged in our analysis. This echoes Nicolaidis and colleagues (64) who found no difference in self-efficacy scores among healthcare providers for caring for their autistic patients by medical specialty, academic degree, or autism training status. One potential reason for this is the possibility that factors such as level of clinical experience or specialty currently make little difference because any autism training received was too generalised or did not target the areas that physicians most require training in, such as how to identify and implement required accommodations. Unigwe and colleagues (61), for example, showed that although autism knowledge was high among their sample of GPs in the UK, perceptions of self-efficacy to manage the ongoing care of autistic patients was low. Further, Tuffrey-Wijne and colleagues (80) found that although HCPs were aware of the need for accommodations for patients with intellectual disabilities, they sometimes struggled to identify what those accommodations were. Therefore, training should incorporate information on common accommodations required within specific settings as a potential means to address this problem.

It is insufficient to focus on physician training alone, however. In the current study, a limited number of potential associated variables were examined, all relating to the HCP. Future research needs to consider system-level variables, such as the financial supports, that might predict responses. It is possible that system-level issues prohibit physicians from feeling empowered or supported to implement accommodations to overcome barriers. Tuffrey-Wijne and colleagues’ work highlighted organisational/system level barriers to adjusting care for people with intellectual disabilities that included a lack of clear lines of responsibility, and a lack of funding and resources for implementing accommodations (80). Following further validation work, it is hoped that the current tool could be used to identify the specific systemic barriers that physicians across specialities and settings experience in relation to treating their autistic patients. It is hoped that this knowledge would guide how to best use the limited resources available in overcoming such barriers by highlighting the specific resources that would be most beneficial in different contexts; for example, funding for a multisensory room may be better allocated to a busy emergency department than a quiet out-patient service which can already provide a quiet area for an autistic individual to wait in until their appointment.

Limitations

This study had a number of limitations which must be taken into consideration when interpreting the results. First, although the sample size was acceptable for an EFA, it was still relatively small. Tabachnick & Fidell recommend a sample size of ~300 in order to be 'comfortable' (35). However, MacCallum and colleagues (81) suggest that sample sizes in the range of 100-200 are acceptable as long communalities are mostly in the 0.5 range and there are is high overdetermination of factors (e.g., at least six or seven indicators per factor and a small number of factors), as was the case in the current study. Regardless, a more stable factor solution may have been obtained with a larger sample size. Future research should investigate this.

A second limitation is the heterogeneity of the sample. It would be expected that physicians from different specialities would experience different barriers to different extents. However, with a lack of sufficient empirical research focused specifically on this to date (23), the tool was designed as a general measure for physicians of all specialities with the hope that this would allow comparisons to be made between specialities, to guide the development of more targeted tools in future. A further limitation related to the sample is that the majority of participants did not have regular contact with autistic patients, although similar rates of contact have been observed in existing literature (32, 33). Due to the lack of specialist autism clinics within physical healthcare in Ireland, it was difficult to identify a specific group of physicians who would have high levels of contact with autistic patients. Therefore, efforts were made to recruit as large and varied sample as possible, in the hope that participants experienced in treating autistic patients would be identified. It is unclear why so many participants did not have much contact with autistic patients. It could be due to non-disclosure of the diagnosis in some cases (71, 74). Low levels of contact do not necessarily mean low levels of knowledge, however, as the participants in the current sample scored highly on the Knowledge of Autism test. Further, all participants had some level of experience with autistic patients, and 60% of the current sample had more than five years' clinical experience. Experience builds over time, so, although high levels of monthly contact with autistic patients was not observed, the participants still had valuable insight to offer. Nonetheless, future research administering the tool to a sample of physicians who are known to have a lot of experience with autistic patients is recommended.

A third limitation is that, although the tool was informed by previous qualitative and quantitative research involving autistic individuals, their supporters, and HCPs as synthesised within a systematic review (23), there was no direct involvement of the autistic community or physicians in the development of the tool. The authors recognise, however, that as research on the barriers to healthcare experienced by autistic individuals increases and tools are developed to represent the perspectives of patients, caregivers and HCPs, there is a need to engage in co-creation of tools and interventions with autistic adults, HCPs, and caregivers (82, 83). Doing so will help to ensure that research priorities are aligned with the autistic community's priorities (84). Future validation work is recommended on the current tool which could include methods such as cognitive interviewing with autistic individuals and physicians to assess the validity of the items from their perspectives.

A fourth limitation is related to potential issues with the face validity of some items within the tool given the unexpected manner in which some items loaded. However, the discrepancies between how some items loaded and the organisation of the taxonomy have potentially highlighted an area of interest regarding how physicians perceive certain barriers, and this warrants further investigation.

A fifth limitation was that, although respondents were asked to report on the severity of the barriers endorsed (data not presented herein) as well as frequency, many respondents did not. It is likely that this was an artefact of how this scale was presented in the questionnaire (i.e., the frequency and severity scales were presented adjacent to one another; Appendix 3.4). As there was so much missing data for this scale, this information was not analysed. Future research should conduct a CFA to examine the items of the current tool with the severity scale as severity is potentially a more important indicator than frequency in the assessment of barriers. A barrier that occurs frequently may not actually pose that much of an issue, however, a barrier that is perceived as severe, whether it occurs frequently or not, could have a much greater impact on the accessibility or experience of care and may therefore be more important to target within interventions. For example, lengthy waiting room times may occur very frequently, but do not pose significant issues for all autistic individuals. It would therefore be more helpful to know how severe this issue is for a particular individual before deciding whether to allocate resources to address this barrier for that individual. Examining the perceived frequency and severity of barriers in tandem could, therefore, offer a greater insight into the experiences of these barriers and therefore be helpful in guiding the prioritisation of attention and resources.

Finally, this paper describes the preliminary assessment of the tool, among an Irish cohort of physicians, only. Due to single administration of the tool, it was not possible to assess test-retest reliability. As there is a lack of fully validated similar physician-report tools for assessing barriers to providing care to autistic individuals, it was not possible to assess convergent/discriminant validity with other measures.

Future research

It is hoped that this tool will eventually be used by physicians to identify the barriers they most commonly experience within their own contexts and use this to generate discussion about accommodations that might be required by their autistic patients. However, it is important to note that the current paper only describes the initial development and evaluation of the tool; further validation work is required. First of all, cognitive interviews should be conducted with physicians to assess the face and content validity of the items within the scale and to assess the presentation and interpretability of the response options. This should than be followed by further psychometric evaluations on the tool: A CFA should be conducted to assess the goodness of fit of the model (39), followed by further evaluations to examine: reproducibility (i.e., does the same factor structure results from the analysis of another sample of responses?); responsiveness (is the tool sensitive to changes?); interpretability (can qualitative meaning be assigned to the quantitative scores?; 39); and usability (i.e., does the tool achieve the specified goals with effectiveness, efficiency, and satisfaction, in the specified context, for the specified end users (40)? Future research should also assess whether adaptations are required to suit different medical specialities (e.g., GPs vs surgeons), or healthcare settings (e.g., primary vs secondary care) and whether these might increase the ‘actionability’ of the resulting data. Future research could also assess whether the tool could be used as a means of benchmarking across specialties and settings and whether benchmarking considerations differ between different specialities. Finally, future work would also need to evaluate the tool in international populations in order to support its use in other countries and healthcare systems.

Future research should also use the current tool with other HCP populations such as nurses and allied health professionals. In a previous study (31), the authors developed a caregiver-report Barriers to Healthcare tool, and Raymaker and colleagues (26) have developed a self-report tool for autistic adults. There is a need however, to involve healthcare providers other than physicians in this type of work as these professionals are also highly involved in the care of autistic patients. Future work may want to use the current tool to assess whether the barriers they experience are different and whether tailored tools are required. Triangulation of this data with the data from the current study and other research with caregivers and autistic adults would provide a truly holistic view of the issues related to autism and healthcare.

In order to further improve the triangulation of data, other methods of assessing barriers are required. Although quantitative methods provide valuable insight, they are unlikely to produce a full understanding the underlying processes (85). Qualitative approaches are, therefore, also recommended to gain a deeper understanding of how and why barriers manifest, as well as to identify potential solutions (86). Patient narratives, which may be gathered through semi-structured interviews, are a recognised means of informing quality improvement initiatives in healthcare (86, 87) and so, should be explored by future research in order to complement and enhance the data gathered by quantitative measurement tools.

Finally, as tools to assess barriers to healthcare for autistic individuals are developed and implemented, there is a need to consider the next step: how to actually implement changes to address the information collected. There are, therefore, two key considerations for future research.

First, there is a need to identify and evaluate the evidence for interventions that have been trialled to date such as autism specific care plans (88) or online autism training for HCPs (89). Second, there is a need for engagement with stakeholders (i.e., HCPs, autistic people, caregivers) to consider mapping of these interventions to the identified barriers and also to consider how to address barriers which are not clearly covered by existing interventions. There now exist a number of good measurement tools, therefore it is imperative that we move our focus to facilitating the use of the valid and reliable data collected by these tools to actually addressing the issues identified and improving the quality of care of autistic patients.

Conclusion

Best practice denotes that physicians and other HCPs provide accommodations to their autistic patients to ensure healthcare is accessible and equitable. The current paper has presented a preliminary version of a novel physician-report Barriers to Providing Healthcare tool which, after further evaluation and validation work, may be used in practice to help physicians distinguish the barriers that exist for them in specific healthcare contexts. Obtaining this information may help identify the supports physicians need to overcome these barriers, and to identify and implement the required accommodations. Finally, as information is gathered on barriers to healthcare for the autism community, there is a need, going forward, to translate this information into effective quality improvement initiatives regarding the care of autistic patients.

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Chapter 5: Study 4

Using Patient Narratives to Explore Challenging Healthcare Encounters

Using patient narratives to explore challenging healthcare encounters for people on the autism spectrum

Where this fits in with the thesis

Although survey studies are important, they cannot provide the whole story of a complex issue (1, 2, 3). Qualitative research is needed to provide context and richer insights into lived experiences (4). This chapter is intended to capitalise on the advantages of qualitative research in order to further our understanding of how and why barriers to healthcare manifest for individuals on the autism spectrum. Thus, Chapter 5 describes the use of patient narratives to explore barriers that occur within challenging healthcare encounters for autistic individuals and addresses the research question: *what barriers contribute to challenging healthcare encounters for people on the autism spectrum?* Data in the form of patient narratives have been shown to contain actionable data for informing quality improvement initiatives in healthcare and thus, the purpose of study 4 was to supplement the quantitative data from Studies 2, 3 and 4 to provide a deeper understanding of how and why barriers occur for people on the autism spectrum and elucidate the impact barriers can have on healthcare experiences.

Peer-reviewed publication

This study is currently under review at a peer-reviewed journal.

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Walsh C, Lydon S, O’Connor P. Using patient narratives to explore challenging healthcare encounters. Under review.

The following chapter is a formatted version of the submitted manuscript to the journal.

Conference presentations

Oral presentation

Walsh C, Lydon S, O’Connor P. Using patient narratives to Explore challenging healthcare encounters. Paper presented at Trinity Health and Education International Research Conference 2021; March 9th-11th, 2021. Virtual event

Author contributions

CW, POC and SL proposed and designed the study. CW collected the data. CW analysed the data and drafted the manuscript. SL and POC conducted secondary analyses of the data and contributed to the manuscript. All authors have read, commented on, and approved the final manuscript.

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Abstract

Introduction

People on the autism spectrum experience health inequities, and often report poor healthcare experiences. Relatedly, autistic people are at risk of a variety of health issues and have a higher mortality rate than non-autistic people. This paper aimed to use patient narratives to systematically explore barriers to healthcare for autistic people and examine how these manifest across healthcare settings.

Methods

Semi-structured interviews were conducted with physicians, autistic adults and caregivers using the Critical Incident Technique. A previously established taxonomy of Barriers to Healthcare for Autistic Persons was used to guide analysis. The severity (i.e., impact on the patient) of described encounters was rated.

Results

A total of 31 participants (*n*=14 physicians; *n*=14 caregivers; *n*=3 autistic adults) describing 46 encounters were included in the final analysis. The most commonly identified barriers to healthcare were associated with autism-related characteristics (e.g., communication/social issues; *n*=40 encounters;87%), followed by healthcare provider (HCP)-related barriers (e.g., poor HCP communication; *n*=39 encounters;84.8%). Physicians most commonly reported barriers associated with autism-related characteristics (*n*=16 encounters, 100%), while caregivers (*n*=22 encounters, 84.6%) and autistic adults (*n*=3;75%) most commonly identified HCP-related barriers. More than a quarter *(n*=12; 26.1%) of encounters were rated as high severity.

Discussion

These data complement quantitative data available through the application of measurement tools and usefully illustrate how barriers manifest within healthcare consultations and the potential impact of poor quality care on the autistic individual. Research must now consider how data on barriers to healthcare access for autistic individuals can best inform interventions to address these issues.

Keywords: Autism; Critical Incident Technique; Health inequities; Healthcare Access; Reasonable Adjustments; Patient Narratives

Conflict of interest

The authors state they have no competing interests.

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Introduction

People on the autism spectrum experience poorer health than the non-autistic population. A plethora of research has now indicated that autistic[4] people tend to experience higher unmet healthcare needs (1, 2), poorer health outcomes (3, 4), poorer health-related quality of life (5, 6) and higher mortality rates than others (7, 8). Further, autistic individuals tend to receive preventative healthcare such as vaccinations (9), or cervical smears (2) at lower rates than others. This is despite autistic individuals presenting to both emergency and non-emergency healthcare services more often (10), being admitted to hospital more often (11), and having longer hospital stays than their non-autistic peers (12). These data are indicative of inequities in healthcare for autistic individuals.

Equity is one domain of healthcare quality (13). Previous research has identified a number of socio-economic determinants of healthcare inequities for people with intellectual and developmental disabilities, including autism, which include factors such as social circumstances (e.g., unemployment); poor communication and health literacy; poor health behaviours; and poor access to quality healthcare (14). Ensuring accessible and equitable healthcare for autistic individuals is now legally mandated in the UK (15, 16). However, a growing body of research has shown that an array of complex barriers to accessing and receiving healthcare still exist for autistic individuals (17-19). These barriers have been shown to exist across the healthcare system: at the level of the patient (e.g., communication difficulties), healthcare provider (e.g., insufficient autism knowledge), and

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[4] Use of identity-first language: many autistic individuals have indicated a preference for the use of identity-first language (i.e., autistic person) as opposed to person first language (i.e., person with autism; 20) Accordingly, this article will use identity-first language.

the system/organisation (e.g., a lack of resources; (17). Given the poorer health outcomes and care experiences of autistic individuals, there is an urgent need to address such barriers (2, 21). Indeed, improving physical health and physical healthcare services is something that the autism community has highlighted as a research priority (22).

Creating change within, and improving, healthcare services requires that healthcare professionals, and healthcare managers, are equipped with the tools, knowledge, and resources they need to care appropriately for autistic individuals (23). A number of survey instruments (e.g., 21, 24, 25) exist to help HCPs and the autism community identify the barriers that exist in healthcare contexts. However, there are limitations to the use of surveys alone for gathering such information. For example, closed questions do not allow patients to present additional or clarifying information. Therefore, the quality of the gathered data may be weakened (26-28). Patient narratives are one means of gathering richer contextual data which may provide these deeper insights (29).

Patient narratives (defined for the purpose of this paper as: “a story of an event or sequence of events told from the perspective of the speaker; 30) have been increasingly elicited by healthcare organisations, providers and advocacy groups as a means of informing quality improvement initiatives in healthcare (31, 32). Narratives are also useful for identifying barriers in various aspects of healthcare, such as medication adherence (33), and early cancer detection (34). Narratives allow the patient, caregivers, or HCPs to describe events in their own words, and provide rich contextual data which has been shown to enhance the patient-centredness of healthcare (31, 35, 36). Examining the narratives of patients, caregivers and HCPs is important because these groups are intrinsically linked within healthcare (37). Therefore, to understand the patient experience, the broader social/and or professional narratives of those who are involved in the patient’s care also need to be understood (38).

The aim of this paper was to employ patient narratives to systematically explore the barriers to healthcare that exist for autistic individuals and how these manifest across various healthcare settings. The specific objectives were: 1) to collect accounts of negative healthcare encounters as reported by physicians, caregivers, and autistic adults; 2) to explore the barriers to healthcare present in these encounters; 3) to assess differences and similarities in contributory factors across groups (i.e., physicians, caregivers, autistic adults; and 4) to assess the severity of these encounters for the autistic individual in terms of their potential to impact on safety of care.

Methods

Design

This study used an exploratory qualitative narrative design using a critical incident technique (CIT) methodology. This study was conducted and reported in accordance with the Standard for Reporting Qualitative Research (39). This design allowed for the elicitation of patients' complete and rich descriptions of challenging healthcare encounters.

Context

Interviews were completed between January 2019-April 2020. The interviews were conducted either in person in the first author's office at the National University of Ireland Galway (*n*=9), via telephone (*n*=36), or video conference (*n*=3). All interviews were audio recorded.

Participants and recruitment

Eligible participants were: 1) parents or caregivers of autistic adults or children living in the Republic of Ireland; 2) autistic adults over the age of 18 who could self-report on their experiences; and 3) physicians working in primary, secondary or tertiary care in the Republic of Ireland. A variety of non-probability sampling methods and recruitment strategies were employed. Participation required informed written consent and was not incentivised.

Recruitment of physicians

Physicians who had taken part in an earlier questionnaire study were given the opportunity to participate in a follow-up interview (25). Advertisements were placed in local and national newspapers, on local radio, and were posted on social media. Information about the study was shared with GPs at annual meetings, and with junior doctors on training days in the local university hospital. Emails of invitation were circulated among staff in one medical school and one hospital group comprising six hospitals. Information about the study was shared at healthcare conferences. Snowballing methods were also employed whereby participants were asked to share information about the study with others.

Recruitment of caregivers

Caregivers who had taken part in an earlier questionnaire study were given the opportunity to participate in a follow-up interview (24). Advertisements were placed in local and national newspapers, on local radio, and were posted on social media. Information packs about the study were sent to primary and secondary schools that had autism units, and to special schools around the Republic of Ireland; principals were asked to share information about the study with the parents in their schools. Autism parent support groups were contacted by telephone, email and social media and asked to share information about the study with their members. Disability support services and organisations were contacted and asked to share information about the study with caregivers of their residential and day clients. Snowballing methods were employed whereby parents and caregivers who participated were asked to share information about the study with other parents/caregivers.

Recruitment of autistic adults

Autistic individuals who had taken part in an earlier questionnaire study (not yet published) were given the opportunity to participate in a follow-up interview. Advertisements were placed in local and national newspapers, on local radio, and were posted on social media. Fliers were placed in universities. Disability services in various third level institutions in the Republic of Ireland were asked to share information about the study. Autism support groups were contacted via telephone, email, and social medial and asked to share information about the study with their members. Snowballing methods were also used whereby autistic adults who participated were asked to share information about the study with their peers.

Data collection

Critical incident technique methodology

The CIT was used to elicit detailed and rich descriptions of the participants' lived experiences of challenging encounters in healthcare and to explore barriers to care as they manifest within such encounters. The CIT interview is a type of cognitive interview used to identify tacit knowledge about a specific encounter (40). It is a flexible method which can be adapted to various researchers and settings (41). Participants are asked to describe a specific event and the interviewer works to enrich the initial summary provided by eliciting further information and details through prompts. The CIT uses a process rather than a strict interview schedule. Table 1 offers an overview of the four 'sweeps' that constitute a CIT interview. The probing questions were based on a previously established taxonomy (17). Some examples of these questions are presented in Table 1. The interviewer, a female PhD candidate, participated in a CIT training session with a senior expert in the technique prior to beginning the study and conducted two pilot interviews with non-academic acquaintances.

Interview process

Prior to the interview, participants were given instruction to guide their selection of an appropriate challenging encounter (See Table 1). It was important that participants were reminded that the encounter needed to relate to physical healthcare as the barriers to physical and mental healthcare likely differ (42). Participants were also cautioned that they should avoid mentioning names in their description. The interviewee proceeded to describe the challenging healthcare encounter they had in mind. Although the interviews were audio recorded, the interviewer also made written field notes relating to the encounter. These notes were relayed back to the interviewee once they had finished their account, for further clarification, correction, or to allow the participant to elaborate further. This process allowed events to be placed in chronological order and repetitions to be omitted. These field notes also offered a useful means of generating effective probing questions. Interviews were gathered until no new themes or categories emerged from interviews and it was judged that data saturation had been achieved.

Table 1. Stages of the critical incident technique interview; adapted from O'Connor et al. (43)

Sweep	Description
Prior to interview	Participant information sheets read: *'prior to the interview, you will be asked to think about a specific physical healthcare encounter which you found frustrating/challenging or in which you felt substandard care was received/you felt you were unable to provide quality healthcare*'. These instructions were repeated before the interview began.
Sweep 1: Description of incident	Participants are prompted to identify and articulate a challenging healthcare encounter they had experienced. Each participant was asked to describe the event from their own perspective in detail, stage by stage, as it had developed.
Sweep 2: Filling in gaps in the incident	The interviewer summarises the incident back to the participant to check the interviewer's understanding. This allowed the interviewer to pinpoint any gaps in time and events, and gave the participant the chance to elaborate on points if they so wished.
Sweep 3: Expanding on the incident	The interviewer reviews the event again, but this time probing at various points and asking for more detailed descriptions of factors that contributed to the challenging encounter. Some examples of

	probing questions included: ‘can you describe what the waiting area was like that day?’; ‘ How did the staff interact with you/your child that day?’; Can you describe anything that help/hindered your communication with the patient?’
Sweep 4: Posing ‘what if’ queries	The interviewer asks questions about the participant’s judgements, thoughts and actions and what would have happened if aspects of the scenario had been different. These questions are designed to obtain additional relevant information and gain a better understanding of the story as a whole. Examples of typical questions were: ‘What if you (doctor) had had more time that day?’; ‘What if the waiting room had been quieter that day’?

Data processing

All recordings were transcribed verbatim and then deleted. The transcripts and field notes were then used to develop a single, rich description of the event or the ‘story’ described in each interview. The interview descriptions were edited into a standard format that was concise, clear, and comparable across the interviews for content analysis. This approach is typical of how CIT interviews are transcribed (40, 43). The transcripts were then assessed for eligibility.

Eligible challenging healthcare encounters had to: 1) describe an encounter relating to physical healthcare; 2) describe a challenging healthcare encounter; 3) describe a specific event or series of events which involved the provision of healthcare for an autistic individual. Ineligible encounters described: 1) encounters pertaining to healthcare other than physical (e.g., mental healthcare; behavioural services; allied health such as occupational or speech and language therapy); 2) a solely positive healthcare encounter; 3) an encounter not focused on one specific care experience or one specific patient.

Content analysis

A deductive content analysis was undertaken (44). Content analysis is a method used to analyse written, verbal, or visual communications (45), the aim of which is to obtain a condensed and broad description of, and produce concepts or categories describing, a phenomenon (44). The content analysis proceeded through the following steps guided by Elo & Kyngas (44):

Step 1. Select the unit of analysis – units of meaning (i.e., statements/phrases concerned with the delivery or accessibility of healthcare) were chosen as units of analysis. Only manifest content was analysed (44).

Step 2. Become immersed in the data – the researcher immersed herself in the data by reading the ‘stories’ many times, considering the questions:

'who is telling? Where is it happening? When did it happen? What is happening? Why is it happening?' (44).

Step 3. Develop a categorisation matrix - A structured categorisation matrix was developed, guided by a previously established taxonomy of barriers to healthcare for autistic individuals (17).

Step 4. Code data according to the categories - All of the data were reviewed for content and coded for correspondence with, or exemplification of, the identified categories (46). Only aspects that fit the matrix of analysis were chosen from the data (47, 48).

To ensure openness and trustworthiness of the coding process, the research team initially independently coded five randomly chosen transcripts. The team then discussed their analyses to ensure consensus was reached about how barriers were coded. All remaining transcripts were subsequently double coded to ensure the coders agreed with the data labels and decisions made to produce those labels (49). No software was used to support the analysis. For any disagreements recorded, the researchers discussed the encounter in question until consensus was achieved. An audit trail recorded decisions made throughout the coding process to enhance dependability and confirmability of the results and to allow the researchers to reflect on decisions. In order to ensure that classifications were adequately internally homogenous and externally heterogenous, the codes applied were supported with examples from interview data.

Rating of severity

The coding of severity (i.e., the potential impact the encounter had on the patient) was informed by the Healthcare Complaints Analysis Tool (HCAT; (50) and the Healthcare Complaints Analysis Tool – General Practice (HCAT-GP; 51) to ensure the severity of encounters across both primary and secondary care could be appropriately categorised. These tools offer guidance on categorising encounters as low, medium, or high severity according to potential impact on the patient. Every encounter was coded by two authors who discussed the data until consensus was achieved.

Establishing trustworthiness

The good practice principles of Lincoln and Guba (52) were applied throughout this study to ensure the rigour of data collection and analysis. *Credibility* (i.e., the value and believability of the findings) was enhanced by: 1) choosing participants with various experiences and perspectives (49); 2) member checking after transcription and prior to analysis, by sending 10 randomly selected participants a copy of their condensed narrative to review for accuracy and to allow participants to express any queries, or request amendments (no participant had any concerns or suggested any changes (52); and 3) double coding the data, and ensuring ongoing open dialogue between the research team regarding coding and analysis. *Confirmability* (i.e., the neutrality and accuracy of the data; 53) was strengthened through continuous engagement by the principal researcher in a systematic reflexive process (i.e., the continuous process of reflection involving the recognition of their situatedness within the research and the effect this may have on the collection and interpretation of the data) throughout the research process (54). *Dependability* (i.e., the reliability of the data; 49) was enhanced through the presentation of the participants' own words through verbatim quotes against the categories they were mapped onto. To facilitate *Transferability* (i.e., whether or not the findings can be transferred into a different situation or context; 53) efforts have been made within this paper to provide rich descriptions of the context, settings, participants, and findings, supported by direct quotations from participants (55). The principal researcher was a female PhD candidate conducting a PhD in the Discipline of General Practice. The primary investigator has a Bachelor's degree in Psychology and Master's degree in Health Psychology. The researcher has previous volunteer experience working with autistic individuals and individuals with intellectual disability but would be classed as an ‘outsider’ to all three participant groups (i.e., autistic individuals, caregivers, physicians).

Ethical approval

Ethical approval was obtained from NUIG Research Ethics Committee (Ref: 17-Nov-20).

Results

Participants

A total of 48 participants were recruited (physicians: *n*=18; autistic adults: *n*=8; caregivers: *n*=22). However, 17 participants were excluded from further analysis because their descriptions of challenging encounters: did not focus on physical healthcare (*n*=13); did not describe a specific event or patient (*n*=2); or were not directly related to autism (*n*=2). As a result, 31 participants who provided narratives of 46 eligible descriptions of challenging healthcare encounters were included in this study. Some participants described more than one encounter. More information on the included participants is provided in Table 2.

Table 2. Participant characteristics

	N **(%)**
Autistic adults (described 4 challenging healthcare encounters)	**3(9.7)***
Age in years (*M; SD)*	19.7 years; *SD*=1
Gender	
Female	3 (100)
Location of healthcare setting	
Urban	1 (25)
Rural	3 (75)
Caregivers (described 26 challenging healthcare encounters)	**14 (45.2)***
Relationship to autistic individual	
Mothers	13 (92.9)
Fathers	1 (7.1)
Gender of autistic individual in caregiver-reported encounters	
Female	1 (7.1)
Male	13 (92.9)
Age in years of autistic individual in caregiver-reported encounters	
< 5 years (preschool)	1 (7.1)
5-12 years (Primary school age)	7 (50)
13-18 years (Secondary school age)	3 (21.4)
>18 years (adult)	3 (21.4)
Location of healthcare setting in caregiver-reported encounters	
Rural	6 (23.1)**
Urban	20 (76.9)**
Physicians (described 16 challenging healthcare encounters)	**14 (45.2)***
Gender	
Female	11 (78.6)
Male	3 (21.4)
Location of healthcare setting	
Urban	13 (92.9)
Rural	1 (7.1)
Specialty/level of seniority	
Junior doctor	1 (7.1)
NCHD	4 (28.6)
Psychiatrist	1 (7.1)
Consultant	1 (7.1)
GP	7 (50)
Years in Clinical Practice	
<5	3 (21.4)
5-10	3 (21.4)
11-20	5 (35.7)
21-30	1 (7.1)
>30	2 (14.3)

*percentage of all 31 participants; **percentage of 26 caregiver encounters; GP=general practitioner; NCHD=non-consultant hospital doctor;

Setting of described negative healthcare encounters

The analysed challenging healthcare encounters occurred in hospital (*n*=28; 60.9%), and primary care (*n*=18; 39.1%) settings. The mean duration of the analysed interviews was 24.58 mins (*SD*=9.06).

Content analysis

The content analysis was structured using a taxonomy of Barriers to Healthcare for Autistic Individuals and barriers are reported across the four central themes of this taxonomy: autism-related characteristics; other patient-related factors; HCP-related barriers; system-related barriers (17). No adaptions to the taxonomy were required based on the analysis.

Table 3. Barriers identified in challenging healthcare encounters reported by physicians, caregivers, and autistic adults with illustrative examples.

Themes and Subthemes	**Identified Examples**
Autism-related characteristics	
Sensory sensitivities - Physician-reported encounters - 31.3% (*n*=5); - Caregiver-reported encounters - 38.5% (*n*=10); - Encounters reported by autistic adults - 25% (*n*=1); - All encounters- 34.8% (*n*=16);	- “She *went berserk because I think it was the fact of the lights, the smell and the whole clinical nature and all the people that were waiting, she went into complete meltdown”* – Caregiver 7. - *“Trying to perform the clinical exam was very tricky; obviously some kids with ASD have sensory and tactile issues and I think for this little lad, those were the main things that seemed to bother him”* – Physician 8 - *“I just really don’t like anyone touching my arm”* – Autistic adult 3.
Communication/social issues - Physician-reported encounters – 75% (*n*=12); - Caregiver-reported encounters – 19.2% (*n*=5); - Encounters reported by autistic adults – 50% (*n*=2); - All encounters - 41.3% (*n*=19);	- *“Sometimes I find it hard to properly explain issues, so that maybe could have contributed, maybe they could have asked better questions to help me along”* – Autistic adult 1 - *“She wasn’t really able to tell us fully what was actually going on with her. You’d ask her about it and she’d say ’oh maybe’, but you couldn’t really get a history out of her*” – Physician 11 - *“You go to the doctor and they go to you ‘what’s wrong with him?’ and I’m saying ‘I don’t know what’s wrong with him, he can’t tell me’ and the doctor is saying like ‘do I have to do a full test, check his liver and everything’ and I’m like ‘well, yeah, because I don’t know”* – Caregiver 13
Issues with waiting - Physician-reported encounters – 12.5% (*n*=2); - Caregiver-reported encounters – 34.6% (*n*=9); - Encounters reported by autistic adults – 25% (*n*=1); - All encounters – 26.1% (*n*=12); -	- *“I like things to be on time, and I’m often left waiting a long time to go in. I just wish that if they know someone with autism is coming in that they’d try to stick to the appointment times they give”* – Autistic adult 3. - *“One day we were waiting for two hours. And I have stressed it before when appointments have come out that he’s autistic and he doesn’t like waiting”* – Caregiver 3 - *“When we do have people waiting in the waiting room , that’s not going to wash either, so it creates all sorts of anxieties”* – Physician 9
Anxiety/other emotions - Physician-reported encounters – 56.3% (*n*=9); - Caregiver-reported encounters – 30.8% (*n*=8) - Encounters reported by autistic adults – n/a	- *“He has a huge fear of needles now”* - Caregiver 3 - *“So we wanted to get scans for her but the problem was, the poor lady was too afraid to get into the CT or the MRI scanner so she started crying her eyes out when we tried to explain it to her”* – Physician 11

- All encounters – 37% (*n*=17);	- *“I always felt very nervous and scared going in to these appointments”* – Autistic adult 1.
Executive function issues - Physician-reported encounters – 6.3% (*n*=1) - Caregiver-reported encounters – n/a; - Encounters reported by autistic adults – n/a; - All encounters – 2.2% (*n*=1)	- *“They (public health nurses) had felt like they were just reaching a wall with him because they would come in, they would tell him you need to do x, y and z and but he wouldn’t be able to mobilise or stop this dysfunctional pattern of not taking care of himself”* - Physician 6
Need for consistency/familiarity - Physician-reported encounters – 50% (*n*=8); - Caregiver-reported encounters – 11.5% (*n*=3); - Encounters reported by autistic adults - n/a; - All encounters – 23.9 (*n*=11);	- *“He tolerated any change in his routine very poorly, any change from his home very poorly”* – Physician 4 - *“And we were just left with this feeling of oh my God, we’ve to start all over again, we have to go through every detail with a brand new person, in a different building”* - Caregiver 14
Behavioural issues - Physician-reported encounters – 37.5% (*n*=6); - Caregiver-reported encounters – 26.9% (*n*=7); - Encounters reported by autistic adults – n/a; - All encounters – 28.3% (*n*=13); -	- *“He was literally kicking and screaming, he was extremely challenging…he would lash out at the team who were trying to help him”*– Physician 4 - *“He really acts up when he’s in there”*- Caregiver 3 - *“She (mother) doesn’t even bring him into the appointments because his behaviour is so difficult apparently”* – Physician 17
Patient-related factors	
Complexity of family involvement - Physician-reported encounters – 25% (*n*=4); - Caregiver-reported encounters – n/a; - Encounters reported by autistic adults – 25% (*n*=1); - All encounters – 10.9% (*n*=5);	- *“He was 20 or 21 and his mum was making a decision like that for him – which probably isn’t inappropriate, but that third person in the room – if she hadn’t brought up that issue, it would have been done weeks earlier than it was and his treatment would have started earlier”* - Physician 13 - *At one point, I was getting a breast exam but my mother was in the room so I wasn’t that comfortable with getting a breast exam right there…but the GP was like ‘oh no, your mother should be here’ and I was confused because I was 18…they looked at my mother for an answer rather than me… so it’s a mixture of my mother being over protective and the GP just being more willing to go with the parent. I am over 18 so I should be able to go to appointments on my own.”* – Autistic adult 2
Healthcare provider-level factors	
Lack of knowledge/skills - Physician-reported encounters – 43.8% (*n*=7);	- *“It was almost like they had never heard what an autistic child was, or they didn’t care, it was just awful”* – Caregiver 4

- Caregiver-reported encounters – 46.2% (*n*=12); - Encounters reported by autistic adults -75% (*n*=3); - All encounters – 47.96% (*n*=22);	- *“I think it was a lack of awareness among staff”* – Physician 3 - *“so I feel like they (doctors) need to learn more about the difference (between genders) and how autism can affect that differently”* – Autistic adult 1
HCP inflexibility - Physician-reported encounters – 12.5% (*n*=2); - Caregiver-reported encounters – 26.9% (*n*=7); - Encounters reported by autistic adults – 25% (*n*=1); - All encounters – 21.7 (*n*=10);	- *“They obviously had a routine of how to do things and there was a rigidity to it which meant that they weren’t able to adapt in order to fulfil what needed to be done”* – Caregiver 6 - *“He’s a very good doctor but he’s very direct, and there’s not really any flexibility so certain patients don’t really go well with that…both the consultant and the patient were very fixed in their ideas of what’s right and what needs to happen…so there was a lack of understanding there”* – Physician 8
Stigma/negative perceptions - Physician-reported encounters – 12.5% (*n*=2); - Caregiver-reported encounters – 23.1% (*n*=6); - Encounters reported by autistic adults – 25% (*n*=1); - All encounters – 19.6% (*n*=9);	- *“And I tried to explain from the start that he has autism, he has sensory issues and the nurse said ‘oh yeah, we get some of them here alright’ …we get some of them! You know those aliens with autism”* – Caregiver 10 - *“Nobody will see these children and this is the issue”* – Physician 4 - *“The GP was probably the worst person I went to, she was just no big deal about it: ‘you’re not doing this right, you should be doing this more’ – it was very much related to stuff that autistic people stereotypically would struggle with”* – Autistic adult 1
Difficulty interpreting symptoms/behaviours - Physician-reported encounters – 81.3% (*n*=13); - All encounters – 28.3 (*n*=13)	- *“So I end up giving them the anti-biotic just in case because it’s so difficult.”* - Physician 1 - *“So that was one thing that I found quite difficult in that scenario, that the child, as part of his normal activity would frequently pull on his ears but I thought that might be a sign of an ear infection but mum said that was just his normal habits”* – Physician 5
Ignoring patient/caregiver concerns/expertise - Physician-reported encounters – 18.8% (*n*=3); - Caregiver-reported encounters – 46.2% (*n*=12); - Encounters reported by autistic adults -75% (*n*=3); - All encounters – 39.1% (*n*=18);	- *“He (consultant) dismissed everything and told my son he had to go back on the medications he’d been on previously and that was it”* - Caregiver 9 - *“She felt very much that she hadn’t been listened to by a lot of different parties and maybe hadn’t been informed as much as she’d have liked”* – Physician 3 - *“It’s difficult because I want to know the risks of certain side effects, so like one says fainting…like how much of a risk is that? who is that a risk for?...which is something you’d think the GP would tell you, but they don’t…I feel like they just want to move on to the next patient”* – Autistic adult 2

Poor HCP communication/ failure to adapt language - Physician-reported encounters – 6.3 (*n*=1); - Caregiver-reported encounters – 34.6 (*n*=9); - Encounters reported by autistic adults – 50% (*n*=2); - All encounters – 26.1% (*n*=12);	- *"He (consultant) is very direct and there's not really any flexibility…so there was a lack of communication there"* – Physician 6 - *"With my home GP, they just give me my prescription and say what's on there but in kind of difficult language. I'm used to reading difficult language now in college but it's different when its medical language which is a whole other area"* – Autistic adult 2
Healthcare system-level factors	
Lack of support for patients/carers - Physician-reported encounters – 12.5% (*n*=2); - All encounters – 4.3% (*n*=2);	- *"It was just hard to see whether or not he would have supports available for him to continue with the behavioural plan when he got home"* – Physician 6 - *"She lives with her elderly brother who is in his 80s and also has some vague diagnosis of an ID. They get about an hour of home help at the moment but we're trying to get them more because they need it"* – Physician 11
Lack of support for HCPs - Physician-reported encounters – 6.3% (*n*=1); - All encounters – 2.2% (*n*=1); -	- *"He would hit and kick and punch and bite and he was a fully grown boy, he was 80kgs. So a lot of the care team couldn't or wouldn't see him because of that…when you have violent patients, it's very, very difficult"*– Physician 4
Time/resource constraints - Physician-reported encounters – 56.3% (*n*=9); - Caregiver-reported encounters – 19.2% (*n*=5); - Encounters reported by autistic adults – 25% (*n*=1); - All encounters – 32.6% (*n*=15);	- *"My GP is often over-crowded so I feel like they just want to move on to the next patient as much as possible, which I understand but it (answering my questions) would help me a lot"* – Autistic adult 2 - *"But I suppose neurologists for children are quite rare, there was only one in Dublin and the time and one in Cork"* – Caregiver 2 - *"I think staffing was a problem, they're such quick interactions, you know*?" – Physician 3
Lack of continuity of care/collaboration between HCPs/services - Physician-reported encounters – 12.5% (*n*=2); - Caregiver-reported encounters – 11.5% (*n*=3); - All encounters – 10.9% (*n*=5);	- *"There was definitely times where there was confusion over care and saying that he needed respite and neurology getting involved in that and I think he'd been seen by somebody else as well, some speciality and they weren't aware of the other one – it was all very disconnected"* – Physician 3 - *"But there was no referral to a specialist neurologist, even though I requested that several times…that was one unfortunate incident whereby the reluctance of doctors to refer to somebody else…I mean why are they so threatened by a specialist, really?…GPs and paediatricians should be using specialists if they need to use them"* – Caregiver 2

Location issues - Physician-reported encounters – 6.3% (*n*=1); - Caregiver-reported encounters – 3.8% (*n*=1); - All encounters – 4.3% (*n*=2);	- *“We had given him something like vallum (under the doctor’s guidance) before we left the house, but we live an hour and a half to two hours from the hospital so sure it was beginning to wear off him before they were able to see him”* - Caregiver 11 - *“I think it might have been a lot because they had to travel from afar as well…a lot of these kids are referred up for sedation for MRIs because they can’t do them in the peripheral hospitals sometimes”* – Physician 2
Lack of qualified personnel - Physician-reported encounters – 6.3% (*n*=1); - All encounters – 2.2% (*n*=1);	- *“I told the nurses and the doctors in there that he’s autistic and he doesn’t like doing this (blood tests) and I just feel they’re not qualified enough”* – Caregiver 3
Inflexible healthcare system - Physician-reported encounters – 6.3% (*n*=1); - Caregiver-reported encounters – 3.8% (*n*=1); - All encounters – 4.3% (*n*=2);	- *“I’m not looking for preferential treatment or anything but there should probably be a better way, especially for children with disabilities and special needs”* Caregiver 5 - *“So it was just a case where the entire system was very unsuited ,to anybody really, but especially to this boy and it just made the entire process so much more stressful for him and his parents* – Physician 3

HCP=healthcare provider; GP=general practitioner; ID=intellectual disability;

Autism-related characteristics

Table 3 provides detailed information on the barriers identified by each group with illustrative examples. Autism-related characteristics were the barrier that emerged most frequently (87% of all encounters; 100% of physician-reported encounters; 80.8% of caregiver-reported encounters; 75% of encounters reported by autistic individuals). Patient difficulties with communication and social interaction were identified as the most common barriers to providing care by physicians. This was followed by the patient's need for consistency/familiarity, and behavioural issues. For caregivers, the main difficulties were related to sensory sensitives, issues with waiting, and anxiety/other emotions. The most commonly identified barriers by autistic adults included communication/social difficulties, sensory sensitivities, and issues with waiting.

Other Patient-related barriers

Other patient-related barriers appeared in 10.9% (*n*=5) of the 46 reported encounters (25% of physician-reported encounters; 25% of encounters reported by autistic adults). These did not appear in any caregiver-reported encounters. Complexity of family involvement was identified in 25% (*n*=4)of physician-reported encounters and 25% (*n*=1) of encounters reported by autistic adults. Attitudes to conventional medicine was not highlighted as a contributory factor in any encounter. Table 3 provides more detail with illustrative examples.

HCP-related barriers

Table 3 provides more detail on the HCP-related barriers identified by each group. HCP-related barriers occurred in 84.8% (*n*=39) of encounters overall (87.5% of physician-reported encounters; 84.6% of caregiver-reported encounters; 75% of encounters reported by autistic adults). A lack of HCP knowledge/skill, the HCP ignoring patient/caregiver expertise/concerns, and poor HCP communication/failure to adapt language were the most commonly identified barriers in encounters reported by both caregivers and autistic adults. The most commonly reported barriers to providing care for physicians were having difficulty interpreting symptoms/behaviours, and a lack of HCP knowledge/skill.

System-related barriers

As shown in Table 3, system-level barriers appeared in 41.3% (*n*=19) of the 46 reported encounters (62.5% of physician-reported encounters; 30.8% of caregiver encounters; 25% of encounters reported by autistic adults). Time/resource constraints were the most commonly reported barrier in encounters reported by all three groups, and the only barrier reported by autistic adults. A lack of continuity/collaboration between HCPs/services occurred in encounters reported by both physicians and caregivers.

Ratings of severity

The majority of the 46 reported encounters were rated as low severity (*n*=21, 45.7%), 13 encounters (28.3%) were rated as medium severity, and 12 encounters were rated as high severity (26.1%). Low severity ratings were related to non-urgent medical care being delayed (*n*=5), staff not communicating what they were going to do (*n*=1), staff speaking in a condescending manner (*n*=1), staff ignoring/dismissing questions/patient suggestions (*n*=4), a noisy reception area (*n*=1), a slight delay in making diagnosis (*n*=8), and staff ignoring the patient (*n*=1). The following excerpts illustrate some examples of low severity ratings.

> *'Our GP is very critical of Finn's*[5] *weight…telling him he needs to be more active and watch what he eats. But Finn is trying to deal with it, and he is getting more active and watching what he eats and he tries to tell the doctor all of this, but the GP won't listen and will just talk over him and won't let him speak'* – Caregiver 3.

> '*But I think both mum and I came to the conclusion that this was just far too distressing for the kid and we'd give it 24 hours, usually with viral processes, things start improving anyways and if they weren't, we'd bring him back in'*- Physician 8

Medium severity encounters included waiting in the emergency department for hours (*n*=5; 38.5%), medical procedures being delayed (*n*=3; 23.1%), and patient or caregiver anxieties not being addressed (*n*=2; 15.4%), some of which are illustrated in the below excerpts:

> '*He had to go for an x-ray, and we had to wait for hours, and waiting is very difficult, he was screaming and he's going a hundred miles an hour and he wants to see what's this guy at, what's that guy at? He doesn't understand sitting down. And every other person was looking at him. It was very frustrating'*- Caregiver 5.

> '*Whenever I ask my GP at home questions, they just don't know what I'm talking about, language sometimes kind of comes out stilted in me, and they would just be like 'whatever' and just not*

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[5] All names are pseudonymised. Names have been randomly chosen from the most common names listed by the Central Statistics Office in 2020 https://www.cso.ie/en/interactivezone/visualisationtools/babynamesofireland/

> *answer my questions or would just tell me to research it'- Autistic* adult 2

High severity ratings related mostly to staff ignoring severe patient distress (*n*=7, 58.3%), as illustrated in the following excerpts:

> *'The last time, for example, they just came with the needles and the bottles and straight away he just lost it and one of the nurses said 'right, were going to start and we're going to do this fast now'. They didn't prepare him, they didn't speak to him, they didn't explain to him what they were going to do. They held his legs and his arms and his shoulders…they didn't even acknowledge that he was screaming his head off'*- Caregiver 3.

> '*I was going to the doctor a lot with a pain in the same area and every time it was like they just didn't believe what was going on, that the pains weren't real….once the pain was really, really bad and I was literally three days in a row in the doctor's trying to get help to get rid of the pain and eventually I just had to bear it and just let it pas*s'- Autistic adult 1.

> '*And then we got her down to the MRI scanner and the radiographers were having none of her either. She was petrified and she was crying and trying to get up and they (radiographers) were like 'this is an extra thing in our day, we don't have time for this'. I really felt a lot of the staff were just 'we don't have time for this, can you please just get on with it and sort it out'*- Physician 11

Other high severity ratings related to refusal to give an appointment (*n*=1), prescribing incorrect medication (*n*=1), being unable to access specialist care (*n*=1), and a missed medical diagnosis (*n*=1).

Discussion

People on the autism spectrum experience health inequities, and often report poor healthcare experiences. In order to provide insights into the barriers experienced by autistic individuals, narratives from physicians, autistic adults and caregivers were collected and analysed in order to identify barriers that occur in challenging healthcare encounters for people on the autism spectrum. The analysed encounters were rated mainly as low severity, but more than a quarter of encounters were rated as high severity. The data illustrate the nature of barriers that emerge and hamper care of autistic individuals.

Barriers associated with autism-related characteristics and the HCP were most commonly identified in the analysed encounters, supporting previous research (17). A number of interventions have attempted to address barriers associated with autism-related characteristics. However, the focus of these interventions tends to be on supporting the autistic individual to engage with, tolerate or anticipate a medical procedure, rather than addressing the HCP or healthcare system. Examples include behavioural interventions to improve the autistic individual's cooperation with injections (56) or improve compliance with physical examinations (57). There is a pressing need to consider interventions that target factors that relate to the HCP and the system which are also known to act as barriers to care. Targeting HCP and system-related barriers may have an impact on patient-level barriers such as those associated with autism-related characteristics. For example, communication issues may be improved through improving HCP communication skills (36). Further, ensuring a quiet space for autistic individuals to wait in until their appointment could alleviate issues with waiting and anxiety (19). Such interventions will be more resource intensive but are necessary if meaningful improvements in care for individuals across the autistic spectrum and the healthcare system are to be achieved (2). Both HCPs and the autism community have acknowledged the need for provider focused interventions such as improved autism training, and various training programmes have been trialled with mixed results (e.g., 11, 58). It is important, however, not to rely too heavily on HCP training as training alone is unlikely to address many of the systemic issues such as a lack of clear lines of responsibility and a lack of funding and resources, which have been identified by HCPs as barriers to implementing accommodations (59). Therefore, more consideration regarding system/organisational-level reforms are also required. Future research may want to use data gathered through existing tools for measuring barriers to healthcare (e.g., 21, 24) to aid in intervention development to ensure that interventions are targeting barriers at all levels.

Barriers at the system level were the least likely to be reported by all three groups, though physicians were more likely than autistic individuals and caregivers to report barriers at this level. These findings support previous suggestions that patients, families, and HCPs may have difficulties identifying system-level factors that may contribute to barriers within healthcare (26). This may be the result of a lack of understanding of factors which occur as the higher systems level, resulting in a tendency to identify proximal, more readily recognised contributory factors (26, 60). Further differences were observed between the barriers reported by physicians, autistic individuals, and their caregivers, which echoes previous findings (17). Physicians were more likely than caregivers or autistic adults to report autism-related characteristics as barriers, while caregivers and autistic adults were more likely to report HCP-related barriers. This information is valuable as it will allow for the development of more tailored interventions and help ensure that interventions aimed at specific groups actually address the issues that are important to that group. These data demonstrate the value of patient narratives as the richer detail provides context which helps to identify latent or system-level issues as well as differences in how and why barriers manifest across groups.

In addition to identifying latent factors, narratives provide valuable insights for both training and quality improvement initiatives which cannot necessarily be obtained through quantitative methods. For example, survey questions are usually set by researchers or service providers and so may miss issues that are important to the patient; deductive style surveys which present lists and checkboxes do not allow the patient to add any new information, so only the prevalence with which items are endorsed can be analysed (26-28). Further, Huppertz and Smith (61), found that although individuals rated their overall healthcare experience as satisfactory on a survey, many provided negative feedback if given space to add comments, indicating that negative aspects of care were only identified through the narrative data and not in the survey. Therefore, survey measurement tools can only provide a limited perspective; they are helpful for identifying a problem but are less helpful for providing insight into why it exists or how to address it (29, 62).

Limitations

There are a number of limitations to this study. First is the under-representation of the autistic voice within the narratives. Recruiting autistic individuals proved difficult and just eight autistic adults were recruited. Of these, just three shared stories that were deemed eligible for inclusion for analysis. Ineligible stories all related to accessing and receiving mental healthcare. These were deemed ineligible because barriers to mental healthcare are likely different from barriers to physical healthcare due to the difference in the nature of the frequency and content of the respective services (42). Future research should employ patient narratives to investigate barriers to mental healthcare services as this is clearly an important issue for autistic adults. Second, the CIT method could be criticised as participant reports could be potentially biased in nature. However, it has been argued that this method of collecting and analysing patient narratives allows for specific encounters to be analysed with increased scrutiny (63). It is hoped that the inclusion of accounts from the perspectives of patients, caregivers and physicians will offset some bias by providing a fuller picture and allowing comparisons to be made between groups. Finally, there is the also the potential for bias in the reporting and analysing of the data, however, by adhering to a rigorous approach and engaging in a reflexive process throughout, it is hoped that this potential has been sufficiently reduced.

Future research

The findings from the current study have a number of implications for future research. Given that HCPs have often reported that they were unsure of how best to improve healthcare for autistic people, there is a need to consider how HCPs can best be supported to reflect on, and learn from, interactions they have had with autistic individuals during healthcare encounters. Prior research has found that patient narratives contain actionable data which can be used by HCPs to identify strategies for improving patient safety (31, 64, 65). Future work may therefore want to evaluate whether physicians could use the information gathered through autism-specific patient narratives to identify strategies to improve the quality of care for their autistic patients. When used in conjunction with reflective practice, video-based patient narratives from people who have intellectual or developmental disabilities (IDD), including autism, and their caregivers, were shown to improve medical students' self-reported comfort and confidence for interacting with patients with IDD in comparison to a control group who received an introductory lecture on IDD only (36). The patient narrative group also demonstrated higher, though not statistically significant, mean performance scores in various simulated clinical scenarios with patient educators (i.e., people with lived experiences of a condition such as IDD). Future research should, therefore, investigate whether patient narratives can be used in a similar way using autism-specific narratives. Consideration is needed on how to help physicians gather such narratives and best use them to improve the quality of care in their settings.

Another method of assessing performance and quality of healthcare is through clinical vignettes (66). A lack of sufficient autism knowledge, which takes into consideration the heterogeneity of the condition, has been frequently noted as a barrier to quality healthcare by autistic individuals, their caregivers, and HCPs (24, 25). Clinical vignettes have been shown to help HCPs and students identify sociocultural factors affecting health and healthcare (67). A variety of socio-cultural determinants of health and healthcare exist for people with disabilities, including autism (14), therefore future research could assess whether patient narratives could be used in the form of authentic clinical vignettes to help train HCPs to become more sensitive to such factors and enhance their ability to reduce access barriers, and improve quality of care (68, 69). Clinical vignettes based directly on patient narratives, may also help physicians to recognise individual manifestations of autism and reduce the occurrence of stereotyping which can result from categorical knowledge of a condition which may be common among HCPs (67). Additionally, patient narratives could also be used to inform authentic standardised patient (SP) scenarios. A growing body of research has provided support for employing autistic standardised patients (SPs) in the training and assessment of HCPs (36, 70, 71). Creating a bank of patient narratives, which could be drawn upon when designing scenarios, would be a valuable resource for researchers and health profession educators and would ensure high quality scenarios which reflect true experiences (72).

Now that a substantial amount of quantitative and qualitative data on barriers to care exist, there is a need to consider interventions to reduce barriers (17-19). Future research should aim develop an understanding of what types of interventions have already been developed, what barriers they aim to address, and how they are being evaluated. Once we have a good understanding of existing interventions and the gaps that remain in this regard, the next step is to engage stakeholders regarding the mapping of these interventions to specific barriers and assessing their appropriateness and feasibility. Patient narratives are useful for this mapping between interventions and barriers through the context they provide. Issues with waiting, for example, is classed as a patient-level barrier and, at first, might suggest implementing behavioural interventions which target the individual's ability to handle waiting areas and waiting times. However, several participants in the current sample noted that issues with waiting would be best overcome by reducing waiting times. A number of physicians in the current sample commented that they already implement strategies to reduce waiting times for their autistic patients, such as giving them the first appointment in the morning, but this is not feasible in every setting. In the emergency department, for example, more innovative, systemic solutions are likely needed such as administrative 'flagging' or alert systems on patients' notes in order to identify an autistic individual who needs accommodations in this regard (73). Therefore, mapping exercises are required to ensure that interventions aimed at specific barriers are actually addressing that barrier effectively and efficiently.

Future work should also be undertaken to understand the relationship between barriers and physical health and to assess whether reducing barriers to healthcare will have a direct or indirect impact on physical health. There was some evidence in the descriptions of challenging encounters in the current study that suboptimal care had the potential for negative impacts on health. In the broader healthcare research literature, improved satisfaction with primary care has been associated with continuity of care (74) and patient compliance with treatment regimens (75), which may indirectly positively affect patient outcomes. It would, therefore, be useful to determine if improvements in accessibility to care translate into better health for autistic individuals. This knowledge would facilitate an understanding of which barriers are most important with regards to health outcomes and should, therefore, be targeted by intervention (17).

Conclusion

Patient narratives are a useful means of identifying the barriers that occur in challenging healthcare encounters for autistic individuals. There is now a need to begin to use this qualitative data in conjunction with the valid quantitative data that already exists to actually implement the necessary reforms to the healthcare system to improve the equity and accessibility of healthcare for autistic individuals.

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Chapter 6: Study 5
A Systematic Review of Interventions to Improve Healthcare Experiences and Access in Autism

A systematic review of interventions to improve healthcare experiences and access in autism

Where this fits in with the thesis

Chapters 2-5 have provided ample data on the barriers to healthcare experienced by autistic individuals. These chapters provided information on the frequency with which barriers occur, how they manifest, and their impact. The findings presented in the preceding chapters, along with existing research, clearly emphasise the need for action. These data must be used to develop effective quality improvement initiatives and interventions to actually improve care (1, 2). Currently, no systematic review has assessed the evidence for interventions that have been trialled to date with the aim of improving access to, or experiences within, healthcare. Therefore, Chapter 6 addresses the research question: *what interventions have been implemented to improve experiences of, or access to, healthcare for people on the autism spectrum and how are they evaluated?* The purpose of conducting this review was to bridge the gap between understanding the barriers that exist and developing effective interventions. Thus, this study provides guidance on the next steps that need to be taken to improve care for autistic individuals.

Peer-reviewed publication

This study has been accepted in a peer-reviewed journal.

The citation is:

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The following chapter is a formatted version of the submitted manuscript to the journal.

Conference presentations

Poster presentation

Walsh C, Lydon S, Walsh E, O’Connor P. A systematic review of interventions to improve healthcare experience and access in autism. Poster presented at Trinity Health and Education International Research Conference 2021; March 9th-11th, 2021. Dublin (Virtual event).

Author contributions

CW, POC and SL proposed and designed the study. CW developed the search strategy and executed the database searches. CW, SL, POC & EW conducted the database screening. CW, SL, POC & EW conducted the data extraction and quality appraisal. CW analysed and synthesised the data. CW drafted the manuscript. CW, SL, and POC contributed to the manuscript. All authors have read, commented on, and approved the final manuscript.

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Abstract

Introduction

Autistic individuals report barriers to accessing and receiving healthcare, and experience increased morbidity and mortality.

Methods

This systematic review synthesises 31 research studies evaluating interventions implemented to improve the healthcare experiences and/or access of autistic persons.

Results

Interventions were most commonly patient-focused (58.1%), focused on supporting the autistic individual to engage with, tolerate, or anticipate, medical procedures, care, or settings. Fewer studies were provider-focused (48.4%) or organisation-focused (6.5%). Interventions were typically evaluated using measures of reactions (45.2%) or behaviour (48.4%), and outcomes were predominantly positive (80.6%).

Discussion

Further research is imperative and should look to how providers and organisations must change. Future research must be inclusive of the autistic community, must measure what matters, and must offer complete detail on interventions implemented.

Keywords: Autism; Autistic; Healthcare; Health Equity; Quality of Care; Systematic Review

Conflict of interest

The authors state they have no competing interests.

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Introduction

Autism is a neurodevelopmental condition characterised by differences in social and communication skills, along with the presence of repetitive and/or restricted behaviours and interests (1). Autistic[6] individuals are more likely to experience illness or disease than others. Poor health is estimated to be more than five times more likely among autistic adults than among non-autistic adults (2). Autistic individuals have a higher likelihood of experiencing a variety of medical conditions such as autoimmune disease, cardiovascular diseases, neurological conditions, and gastrointestinal disorders (3). A number of studies, analysing data from different countries, have also evidenced substantially increased mortality, in some cases up to twice as high, among autistic persons (4-7). This means autistic individuals are more likely to die than their peers over a period of time. Further, compared to non-autistic controls, autistic individuals have a greater number of unmet healthcare needs (8), receive less preventative care (8), report more frequent use of emergency care (8, 9), and have a lower health-related quality of life (10, 11). However, poor health is not an inevitable consequence of autism, but instead reflects important health inequities experienced by autistic individuals that exist internationally (9, 12).

Health inequities are defined as “unjust and avoidable differences in health care access, quality, and outcomes” (13, p.1; 14). Potential determinants of, or contributors to, the health inequities observed among persons with intellectual disabilities are well-delineated and are suggested to include increased experience of undesirable social determinants of health (e.g., poverty, unemployment), communication problems and health

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[6] We have deliberately opted to use identity-first language (i.e., autistic individual) rather than person-first language (i.e., individual with autism) in this manuscript. The appropriate use of language around autism is recognised as a complex issue (15). However, we have made this decision as, in recent years, autistic individuals have expressed a strong preference for the use of identity-first language (16,17) and the use of person-first language has been suggested to perpetuate or sustain stigma around disability (18).

illiteracy, poor health-related behaviours, and poor access to, and quality of, healthcare (19, 20). It is likely that such factors, and potentially, also play a role in perpetuating health inequities for autistic individuals (21-24) making this a complex issue. For instance, research shows that autistic people are more likely to be less educated, unmarried, and more socially deprived than the general population (21). Similarly, it is well-recognised that autistic individuals experience issues in accessing and receiving high quality healthcare (22-24).

Barriers to healthcare access can occur at the level of the patient, the healthcare provider (HCP), and the healthcare system (24). Key barriers to healthcare access occurring at the level of the patient can include: sensory sensitivity which makes it difficult for the individual to experience novel or stimulating settings or to tolerate physical exams or investigations (22, 24); communication difficulties which complicate identification of pain or symptoms and engagement with HCPs regarding investigations or treatments (24, 22); and behaviours that challenge (e.g., self-injurious behaviour; aggression) which can cause the discontinuation of medical procedures, result in the use of physical and/or chemical restraint (e.g., 25, or make caregivers less likely to attend for medical appointments (24). Barriers to healthcare access occurring at the level of the HCP include: a lack of provider knowledge or skill relating to autism which impedes their ability to adapt care for autistic patients or to understand their experiences or needs (22, 24); inflexibility of HCPs or a lack of willingness to make accommodations or change their behaviour to facilitate the autistic patient (22, 24); and HCPs ignoring, or not taking seriously, the concerns or expertise of the autistic patient or their caregiver (22, 24). Barriers also occur at the level of the organisation or health system and these include: a lack of continuity of care or collaboration between the different HCPs or services involved in the management of the autistic patient’s care (24); time or resource constraints which impede the delivery of high quality care to the autistic patient (24); and financial and/or insurance issues which can preclude autistic individuals from accessing required care or making use of preventative care services (24). Systematic reviews (22, 24) of barriers to healthcare access showcase the range of barriers that can impede care and emphasise the complexity of the challenge of improving healthcare access and experiences for autistic persons.

It has been mandated in the UK that public services adjust their practices to accommodate autistic people (26, 27). Further, the development of autism-friendly healthcare services is a recognised research priority of the autistic community (28). There is an increasing focus internationally on making environments and processes “autism-friendly” which involves identifying the challenges or difficulties that autistic individuals may experience when engaging with them and making adaptations to facilitate the participation of autistic individuals (e.g., addressing sensory sensitivity by reducing noise levels or removing fluorescent light to make the environment less stimulating (29). However, although the barriers to healthcare access have been well-considered and shown to exist across many countries (22-24), there is a lack of guidance on how to improve the care of autistic patients. To-date, suggestions for improving healthcare have been described (24, 30-34) but there has been no attempt to synthesise interventional research in this area and to examine the types of interventions which have been employed and their outcomes. Understanding what type of interventions have been trialled to-date, and their effects, is important for informing both research and practice in this area. Accordingly, the purpose of this systematic review was to synthesise the research evaluating interventions to improve the healthcare experiences and/or access of autistic persons.

Methods

Study Design

This systematic review was conducted, and is reported, in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-analysis (PRISMA) guidelines (35). The protocol was registered in the International Prospective Register of Systematic Reviews (PROSPERO; CRD42020163200).

Search Strategy

A systematic search of five electronic databases was completed in January 2020. The databases searched were: Medline, CINAHL, Web of Science, Psychology and Behavioral Sciences Collection, and PsycInfo. The search protocol, developed with the assistance of a research librarian, included Medical Subject Headings (MeSH) search terms along with other keywords that related to autism, healthcare, and interventions. The search strategy for Medline is presented in Appendix 4.1 and was adapted as necessary for the other databases. Searches were limited to the English language, but no restriction was placed upon year of publication.

It is well recognised that the exclusion of grey literature (i.e., materials not published in peer-reviewed journals) can lead to the over-estimation of intervention effects within systematic reviews (36). Accordingly, grey literature searches were undertaken in November 2019 to identify unpublished materials suitable for inclusion in this review. Searches were conducted across: 1) Google (first 100 returns; location set to UK); 2) Google Scholar (first 100 returns; location set to UK); 3) Ethos (e-theses online service; all returns examined); and 4) OpenGrey (System for Information on Grey Literature in Europe; all returns examined). Across each of these databases, the keyword “autism” was entered along with one of the following terms: health; healthcare; hospital; general practice; doctor; physician; nurse.

In order to identify additional relevant studies, the reference lists of a number of related reviews (37-40) were screened along with the reference lists of all studies determined to be suitable for inclusion following the electronic searches.

Study Selection

Inclusion and Exclusion Criteria

In order to be included in this review, studies were required to: a) be written in the English language; b) describe the implementation and evaluation of an intervention to improve health care access or care experiences for autistic persons in relation to physical healthcare specifically (defined as: any medical care related to the physical wellbeing of the participants; 24); and c) use an established research design. The interventions described could include autistic individuals or their caregivers, healthcare staff, or could target change or improvement within organisations or health systems.

Studies were excluded if they involved the development or consideration of relevant interventions but did not evaluate these (e.g., 41). In addition, studies were excluded if their focus was not specific to autism and/or it was not possible to extract data pertaining to the outcomes of the intervention for autistic participants or their caregivers (e.g., 42). Other reasons for exclusion included: implementation of an intervention with a focus on improving HCPs' diagnostic or screening capabilities (e.g., 43); interventions implemented in mental health or dental settings (e.g., 44); case reports of studies or multiple case studies reported together (e.g., 45); no original, empirical data provided (e.g., 46). In some cases, multiple exclusion criteria were relevant to an individual study.

Screening

Titles and abstracts of all records returned during the electronic searches were screened by one author. If a study appeared relevant, or it was not possible to determine whether it met the inclusion and exclusion criteria, the full text was accessed. Full text review was completed by the full research team in tandem, and decisions regarding inclusion or exclusion were documented.

Data Extraction and Synthesis

Two authors independently completed data extraction with any disagreements resolved through discussion until consensus was achieved. Data were extracted on: country, intervention setting(s), participant(s), research approach, intervention characteristics, evaluation measures, and outcomes.

As the included papers were diverse with regards to research design, and interventions implemented, it was necessary to develop and apply codes to facilitate data synthesis. Throughout the coding process, researchers were careful to ensure that the context and meaning of data was retained in spite of the application of codes (47). Coding was completed by the research team together following completing of initial data extraction, discussing each study in detail prior to developing and/or selecting the appropriate code(s). The importance of dialogue between researchers during coding has been outlined previously (48). Table 1 presents a complete summary of codes applied to synthesise data on participants (e.g., autistic children/adults, HCPs), setting (e.g., educational setting for autistic children/adults, primary care), evaluation measures (e.g., measures of reactions, measures of behaviour), the intervention implemented (e.g., patient-focused, provider-focused), and the outcomes of the intervention (e.g., positive, mixed). With regards to evaluation measures employed, Kirkpatrick’s evaluation hierarchy (49) was used to categorise the level of evaluation completed as has been done in other systematic reviews previously (50, 51). Further, outcome data within each paper were coded using a grading system of positive/mixed/negative/no clear effect, a similar process to that employed in other systematic reviews (e.g., 40) previously. Multiple codes were applied if required to capture the data presented.

Methodological Rigour

Methodological rigour was assessed by two researchers working in tandem and using the Quality Assessment Tool for Studies with Diverse Designs (QATSDD; 52) which allows for the appraisal of studies using quantitative, qualitative, or mixed methods approaches. This 16-item tool has been widely applied in systematic reviews pertaining to autism and/or health services research (24, 40, 53, 54). Items are rated on a four-point scale (0-3), with a higher score indicating greater rigour.

Table 1. Codes applied to summarise data relating to participants, settings, the type of evaluation measures applied, the nature of the intervention, and the outcomes achieved

Participants	Setting	Type of Evaluation Measures Employed	The Nature of the Intervention	Outcomes Achieved
Autistic children/adults Any autistic participant(s), regardless of age or any indicated co-occurring diagnoses or conditions. ---------------------------- *Healthcare providers* Any participant(s) involved in the delivery of medical care to patients, including physicians, nurses, emergency medical services personnel and others. ----------------------------	*Educational setting for autistic children/adults* Any setting(s) which offered educational services for autistic children or adults. ---------------------------- *Secondary care* Any hospital or outpatient settings offering physical healthcare services. ---------------------------- *Primary care* Any community-based healthcare services, to include	Level 1: Measures of *Reactions* Measures relating to the perceived likeability, usefulness or relevance of the intervention implemented. --------------------------------------- Level 2A: Measures of *Learning-Change in Attitudes* Measures which are focused on assessing changes in attitude or perceptions towards constructs targeted by the intervention. --------------------------------------- Level 2B: Measures of *Learning-Changes in Knowledge or Skills*	Interventions which are *patient-focused* Interventions that are focused on supporting the autistic individual to engage with, tolerate, or anticipate, medical procedures, medical care, or healthcare settings. --------------------------------------- Interventions which are *provider-focused* Interventions which are focused on equipping healthcare providers with the knowledge and skills required to	Outcome data were coded as *positive* if data were demonstrative of desirable changes in the outcome measure(s) or suggestive of a positive impact of the intervention. ---------------------------- Outcomes were coded as *negative* when the nature of changes observed on the measure(s) was undesirable or the data

Caregivers of autistic children/adults Any participant(s) identified as caregivers to autistic individuals receive healthcare services, to include family members or those employed as support workers for autistic individuals. ---------------------------- *Healthcare administrators* Any participant(s) involved in the organisation or management of healthcare services or settings. ---------------------------- *Health Profession Students*	general or family practice settings. ---------------------------- *Home and community* The participant's home or other settings in their community or locality. ---------------------------- *Health profession education setting* Any setting or location in which health profession educational content is delivered/taught or assessed. ---------------------------- *Prehospital care* Any setting or location used by emergency medical	Measures which are focused on assessing for demonstrable changes in knowledge or skills related to the intervention. -------------------------------------- Level 3: Measures of *Behaviour* Measures focused on changes in related behaviour or transfer of learning to the clinical setting. -------------------------------------- Level 4A: Measures of *Results: Changes in Organisational Practices* Measures focused on examining the impact of the intervention upon the practice and functioning of the organisation. --------------------------------------	effectively support and care for autistic patients. -------------------------------------- Interventions which are *organisation-focused* Interventions focused on altering the physical environment or organisation in ways that create a more inclusive and supportive environment for autistic individuals.	were suggestive of a negative impact of the intervention ---------------------------- Outcomes were coded as *mixed* in instances where outcome data were suggestive of both positive and negative impacts of the intervention. ---------------------------- Outcomes were coded as *no clear effect* when null results were reported or the research approach did not support determination of the intervention outcome.

Any participant(s) completing a health-profession related educational degree, to include medical, nursing, and other health sciences students.	services, to include ambulances or related training facilities. ---------------------------- *Other or unclear* This code was applied when it was not possible to determine the setting in which the intervention was delivered, or the setting was not related to any of the above codes.	Level 4B: Measures of *Results: Changes in the Organisation and/or among Employees* Measures which assess for improvements in an organisation or health system or for its employees or patients.		

Results

Figure 1 presents a PRISMA flow diagram depicting study selection. Titles and abstracts were examined for more than 3,800 records with full-texts considered for 191 papers and 30 papers (55-84) describing 31 studies, ultimately included. Almost all studies (96.8%) were peer-reviewed with the exception of one study (76) which comprised a student thesis. Of the 31 studies included, more than 70% were conducted in the US and 16% conducted in Canada. Studies conducted outside of North America were much fewer in number (<15% of included studies). Table 2 provides a summary of the characteristics of included studies with a study-by-study summary available in Appendix 4.2.

Participants

As shown in Table 2, participants in the studies were most commonly autistic children or adults (61.3% of studies), followed by HCPs (29% of studies). The participation of caregivers of autistic children or adults, healthcare administrators and health profession students was less frequent (all <15% of studies) across the studies reviewed.

Setting

Studies were most commonly conducted in primary care, secondary care, or educational settings (all in 22.6% of studies) that served autistic children or adults (see Table 2). Studies conducted in participants’ homes or the community, health profession education settings, prehospital care or elsewhere were less frequent.

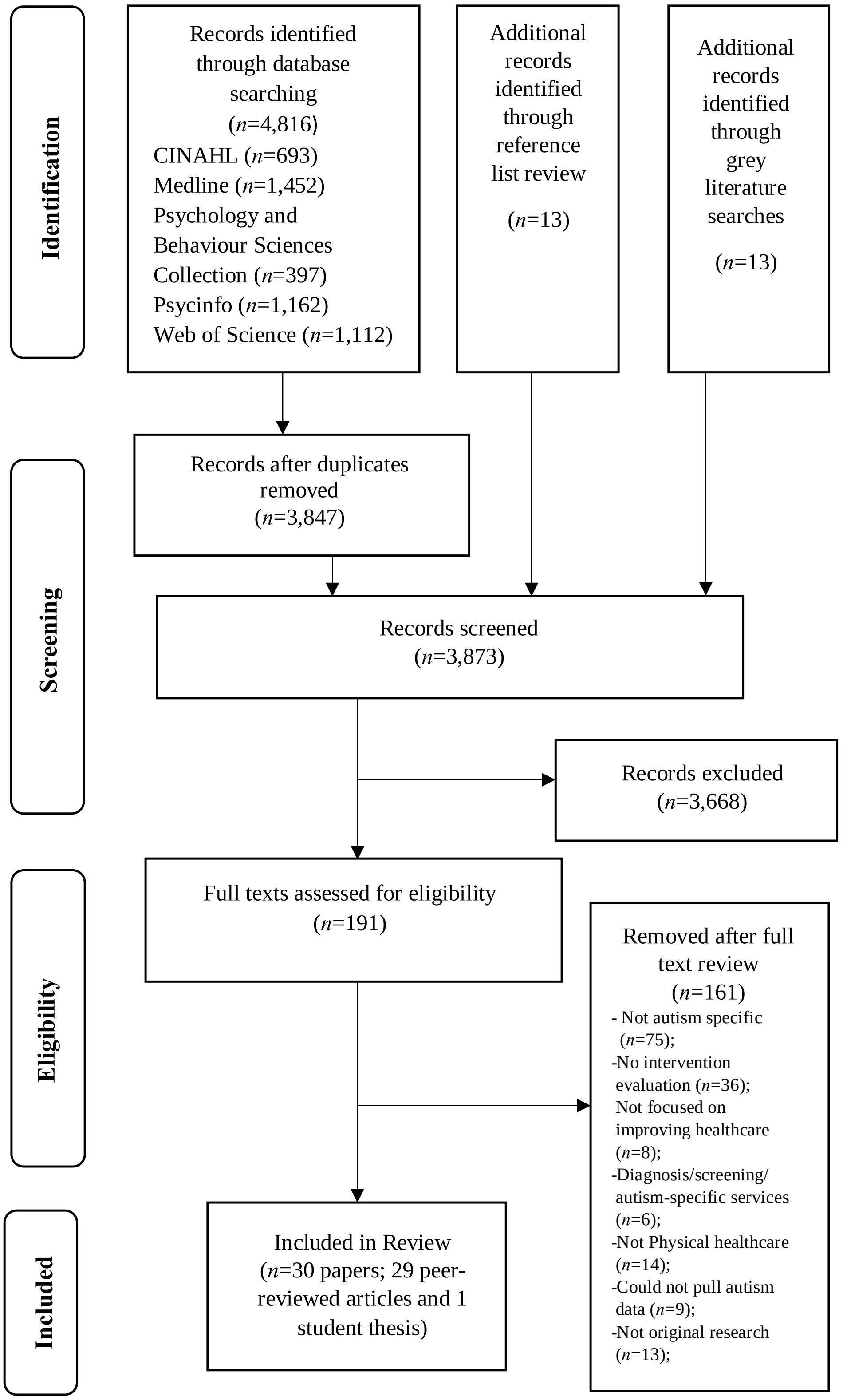

Figure 1. PRISMA flow diagram of study selection

Table 2. Summary of the characteristics of included studies (n=31 studies)

Summary		**N of Studies (%)***
Country	USA	23 (74.2%)
	Canada	5 (16.1%)
	UK	2 (6.5%)
	Other (i.e., Turkey, France)	2 (6.5%)
Participants	Autistic Children/Adults	19 (61.3%)
	Healthcare providers	9 (29%)
	Caregivers of autistic children/adults	4 (12.9%)
	Healthcare administrators	3 (9.7%)
	Health profession students	2 (6.5%)
Setting	Educational setting for autistic children/adults	7 (22.6%)
	Secondary care	7 (22.6%)
	Primary care	7 (22.6%)
	Home and community	5 (16.1%)
	Health profession education setting	4 (12.9%)
	Prehospital care	1 (3.2%)
	Other or Unclear setting	4 (12.9%)
Research Approach	Quantitative- groups research	16 (51.6%)
	Quantitative- single subject research	10 (32.3%)
	Mixed methods	5 (16.1%)

Note. Percentages do not total to 100% as some studies fell within more than one of the categories presented.

Research Approach

As outlined in Table 2, studies were predominantly quantitative only, typically using groups research designs (51.6%; e.g., pretest-posttest design; quasi randomised controlled trial) though single-subject research designs (e.g., changing criterion design; multiple baselines across participants design) were also common (32.3%). Five studies (16.1%) used a mixed methods research approach, collecting both qualitative and quantitative data as part of the intervention evaluation conducted.

Level of evaluation

Full detail on the evaluations of interventions within studies is provided in Appendix 4.2, while Table 3 provides a summary of evaluation across the included studies, examples of evaluations conducted, and associated outcomes. Studies most commonly assessed behaviour (54.8% of studies; e.g., compliance with components of a medical exam) followed by the assessment of reactions (45.2%; e.g., caregiver satisfaction with care) and then changes in attitudes (29%; e.g., confidence working with autistic patients). As can be seen in Table 3, for each level of evaluation, the majority of outcomes observed were positive.

Outcomes

In total, 80.6% (*n*=25) of studies demonstrated positive outcomes. A further four studies showed positive outcomes on some measures and either no clear effects (*n*=3; 9.7%) or mixed effects (*n*=1; 3.2%). The two final studies reported outcome data that was classified as no clear effects.

Table 3. Levels of evaluation across the studies reviewed with examples and related outcomes

Level	Example	*N* studies (%)	Outcomes (*n* studies; %)
1. Reactions *Measures relating to the perceived likeability, usefulness or relevance of the intervention implemented.*	- Survey on ease of completion of educational course, knowledge gained, helpfulness of information provided, and likelihood of recommending course to colleague (78) - Caregiver satisfaction with care measured during a routine postoperative telephone call (82)	14 (45.2%)	- Positive (*n*=14/14; 100%)
2A. Learning: Change in Attitudes *Measures which are focused on assessing changes in attitude or perceptions towards constructs targeted by the intervention.*	- Survey seeking information on subjective comfort with responding to acute crises in autism (74) - Questions relating to understanding challenges of, and confidence and comfort with, working with autistic patients (69)	9 (29%)	- Positive (*n*=6/9; 66.7%); - No clear effects (*n*=2/9; 22.2%); - Mixed (*n*=1/9; 11.1%)
2B. Learning: Change in knowledge/skill *Measures which are focused on assessing for demonstrable changes in knowledge or skills related to the intervention.*	- Survey seeking information on knowledge of autism (74) - Assessment of knowledge about hospital resources for assisting autistic children (71)	4 (12.9%)	- Positive (*n*=3/4; 75%); - No clear effects (*n*=1/4; 25%)
3. Behaviour *Measures focused on changes in related behaviour or transfer of learning to the clinical setting.*	- Head motion steadiness while lying in the mock MRI scanner (61); Study 2) - Compliance with components of the medical exam and problem behaviour (62)	17 (54.8%)	- Positive (*n*=16/17; 94.1%) - No clear effects (*n*=1/17; 5.9%)
4A. Results: Change in system/	- Length of patient stay (56) - Use of sedation (82)	4 (12.9%)	- Positive (*n*=3/4; 75%)

organisational practice *Measures focused on examining the impact of the intervention upon the practice and functioning of the organisation.*				-	No clear effects (*n*=1/4; 25%)
4B. Results: Change among Participants *Measures which assess for improvements in an organisation or health system or for its employees or patients.*	-	Subjective ratings of progress towards achieving family-centred care goals (57)	1 (3.2%)	-	No clear effect (*n*=1/1; 100%);

MRI=magnetic resonance imaging;

Nature of the intervention

For complete information on interventions delivered please see Appendix 4.2. Table 4 provides an overview of the types of interventions reported, along with examples and information on outcomes. A majority of studies were patient-focused (58.1%) with the intervention predicated on supporting the autistic patient to engage with, tolerate, or anticipate, medical procedures, care, or settings. Targets in these studies varied but included teaching cooperation with injections (55) and improving communication with autistic patients during medical exams (76). These interventions typically took the form of a behavioural intervention (72.2%). Outcomes of patient-focused interventions were predominantly positive (88.9% showing positive outcomes only).

Interventions were also commonly provider-focused (48.4%), or intended to equip HCPs with the knowledge and skills required to effectively support and care for autistic patients. Targets within studies varied but it was most common for studies to seek to improve staff knowledge of autism or attitudes towards autistic patients. These interventions were typically educational interventions (66.6%), though a small number of studies did describe care plans or quality improvement deemed to be provider-focused. Outcomes of provider-focused interventions were typically positive (66.6% of studies showing positive outcomes only).

Only two studies (6.5%) reported the use of interventions classified as organisation-focused, and which dealt with altering the physical environment or organisation in ways that created a more inclusive and supportive environment for autistic individuals. Both studies described the use of care plans for autistic patients, with one study documenting positive outcomes only (83) and the other showing a positive effect on two outcome measures and no clear effect on a third (82).

Methodological Rigour

Methodological rigour was variable. QATSDD scores can range from 0 to 42 (Qualitative or Quantitative Studies) or 46 (Mixed Methods Studies). Mean QATSDD score was 18.3 (SD=5.5, range=9-28). Studies performed best on items assessing description of the research setting, aims and objectives, and data collection procedures. Studies performed most poorly on items appraising consideration of sample size, justification of analyses and user involvement in design.

Table 4. Types of interventions reported across the included studies, with examples and related outcomes

Nature of Intervention; n (%)	Type and Examples	Outcomes* (n; %)
Patient-focused; 18 (58.1%)	**Behavioural Intervention** (*n*=13/18; 72.2%) - e.g., Behavioural intervention drawn from science of Applied Behaviour Analysis which comprised of skills teaching, a token economy and reinforcement to teach cooperation with injections (55).	- Positive (13/13; 100%)
	Picture Schedule (*n*=2/18; 11.1%) - e.g., The use of pictures schedules to inform autistic patients of what is expected during a hospital visit (59).	- Positive (2/2; 100%)
	Care Plan (*n*=2/18; 11.1%) - e.g., use of the autism Healthcare Accommodation Tool which allows patients to create a personalised accommodations report for their primary care provider (77).	- Positive (1/2; 50%) - Positive and No Clear Effects (1/2; 50%)
	Quality improvement (*n*=1/18; 5.6%) - e.g., autism-specific medical home intervention that utilised a QI approach and included accomplishments such as an autism care plan, tools to improve appointments, coordination with outside resources, longer duration appointments etc. (68).	- No Clear Effects (1/1; 100%)
Provider-focused; 15 (48.4%)	**Educational Intervention** (*n*=10/15; 66.6%) - e.g., an online training module for healthcare professionals that covered an introduction to autism and recommended proactive and reactive strategies for caring for autistic paediatric patients (78).	- Positive (8/10; 80%) - Positive and Mixed (1/10; 10%) - Positive and No Clear Effects (1/10; 10%)
	Care Plan (*n*=4/15; 26.7%) - e.g., An autism specific care plan was developed and addressed: expressive and receptive communication, social and pragmatic concerns, and safety. It was completed by patient and uploaded to the patient’s electronic record (56).	- Positive (2/4; 50%) - Positive and No Clear Effects (2/4; 50%)
	Quality improvement (*n*=1/15; 6.7%) -e.g., Teams developed plans for change (e.g., improving chronic care management of autistic children) and a timeframe. To support implementation of plans, there were monthly conference call on autism-related topics and two site visits from a QI specialist (57).	- No Clear Effects (1/1; 100%)
Organisation-focused; 2 (6.5%)	**Care Plan** (*n*=2/2; 100%) - e.g., An individualised written perioperative plan for autistic children intended to serve as a management guide and available to all hospital personnel to optimise perioperative patient cooperation and avoid harm. Covered factors including optimal time of day for procedure, modification of hospital arrival time, avoidance of multiple transitions etc. (82).	- Positive (1/2; 50%) - Positive and No Clear Effects (1/2; 50%)

Note. QI=Quality Improvement. *The data resulting from each outcome measure was graded as positive/negative/mixed or no clear effects. Therefore, studies could receive more than one outcome categorisation depending on the number of measures used.

Table 5. Comparison of studies using single-subject research designs and groups research designs

Variable	**Single-subject Research Designs (n=10 studies)** *n*; %	**Groups Research Designs (n=21 studies)** *n*; %
Level of Evaluation		
Level 1- Reactions	1; 9.1%	13; 34.2%
Level 2A- Learning: Change in Attitudes	-	9; 23.7%
Level 2B- Learning: Change in knowledge/skill	-	5; 13.2%
Level 3- Behaviour	10; 90.9%	6; 15.8%
Level 4A- Results: Change in system/organisational practice	-	4; 10.5%
Level 4B- Results: Change among participants	-	1; 2.6%
Nature of the Intervention		
Patient-focused	10; 100%	8; 32%
Provider-focused	-	15; 60%
Organisation-focused	-	2; 8%
Outcomes		
Positive	11; 100%	31; 81.6%
No Clear Effects	-	6; 15.8%
Mixed	-	1; 2.6%
Methodological Rigour		
Mean QATSDD Score	20.1	17.7
Range	13-28	9-28

Note. As described in the method section, it was possible for multiple codes to be applied within the same study. Therefore, the numbers presented herein do not align with the total number of included studies.

Comparison by Research Design

Of the 31 studies included, 11 utilised a single-subject research design (35.5%; e.g., changing criterion design; reversal design) and 21 utilised some form of groups design (67.7%; e.g., randomised controlled pilot trial study). Table 5 presents a comparison of the level of evaluation, nature of intervention, outcomes, and methodological rigour by research design. As can be seen, there appeared to be less variability within the methods and outcomes of studies utilising single-subject research designs.

Discussion

Morbidity and mortality are substantially increased among autistic individuals internationally. Relatedly, autistic individuals experience issues in accessing and receiving high quality healthcare that are not experienced by non-autistic individuals (22-24). There is therefore a need, and in some countries a legal requirement, to develop and implement interventions that can improve the healthcare experience or access of autistic persons. Accordingly, the current systematic review offers a synthesis of research on interventions to improve healthcare experiences and/or access for autistic persons. Key findings include the small body of research describing evaluations of relevant interventions, the predominant focus on changing the behaviour of the patient with comparatively little work focused on changing HCP behaviour and/or healthcare organisations and systems, and relatively weak forms of outcome measurement.

Although many papers exist that provide recommendations for providing healthcare to autistic individuals (30-34), a much smaller body of literature (*n*=30 papers) has evaluated interventions to improve access to, or experiences of, healthcare for autistic persons. Of the studies reviewed, only 13% were conducted outside of North America and just under half (48.3%) took place in a healthcare setting. A recent workshop (28) that brought together autistic people, their relatives, clinicians, healthcare managers and others, identified the development of autism-friendly healthcare services as a research priority. While it is encouraging that a majority of studies reviewed herein (80.6%) demonstrated positive outcomes of interventions, the autistic community is heterogeneous (32) and additional research will be required to explore, and establish, a variety of evidence-based practices suitable for facilitating care of autistic individuals of all ages, and across all health specialties and settings. This is particularly true given that none of the included studies employed a full randomised controlled trial design (two studies employed randomisation but were described as pilot studies) which continues to constitute the ‘gold standard’ for interventional research and to afford the most trustworthy evidence on effectiveness (85, 86). However, research has now effectively delineated the barriers to healthcare access (22, 24, 87) and there exist a number of tools to facilitate data collection in relation to the barriers to healthcare access experienced by autistic adults (8) or the caregivers of autistic persons (24) or HCP (24) which should facilitate future research in this area. This research must move beyond developing understanding on, and collecting data relating to experiences of, healthcare access and experiences of autistic persons to ‘actioning’ data to inform the development and implementation of interventions to improve the care and health of autistic persons.

It is notable that interventions were most commonly categorised as patient-focused (58.1% of studies), and sought to support the autistic individual in engaging with, tolerating or anticipating, medical procedures, care or settings. All studies using single-subject research designs reported on patient-focused interventions which likely reflects the popularity of the design type in behaviour analytic research (88) and the frequency with which behaviour analysts work directly with autistic children and adults (89). It is certainly known that autism-related characteristics (e.g., sensory issues, communication issues, challenging behaviour) are barriers to healthcare access and may impede the provision of high quality care for autistic patients (24). However, autistic individuals and caregivers have also highlighted HCP-related barriers (e.g., lack of knowledge/skills, inflexibility) and healthcare system-related barriers (e.g., lack of continuity/collaboration, lack of time/resources), which were addressed less frequently in the included studies (48.3% of studies and 6.5% of studies respectively). Future research must look beyond approaches targeting only the behaviour of the autistic individual and must instead ensure that interventions target the other factors that are known to impact upon access to, and experiences of, care (24). Although interventions seeking to change HCP behaviour, or management, coordination, or facilities within organisations or healthcare services, may be more complex or resource-intensive, they are essential to ensure that care is improved for the many diverse autistic persons accessing healthcare services daily and addressing unmet needs and issues of poor care which persist in the autistic community (8). Researchers, or quality professionals, may wish to use existing tools to establish the barriers to care in advance of intervention development to ensure that interventions are likely to yield perceptible improvements for autistic patients. For example, Raymaker and colleagues' (90) have developed a tool that allows autistic individuals to self-report barriers experienced in healthcare settings, Walsh et al. (91) present a tool which allows the caregivers of autistic patients to report on the frequency and severity of barriers experienced in physical healthcare settings, and Walsh et al. (92) have completed initial validation of a tool which allows HCPs to identify barriers to care provision or access for autistic patients. The use of such tools will allow providers or organisations to determine the most frequent (and in some instances, the most severe) barriers impacting care for autistic patients within their organisation or setting. This may allow prioritisation and evaluation of interventions or strategies which are likely to address these specific barriers. Walsh and colleagues (24) have offered recommendations for interventions to address the most common barriers to healthcare access for autistic patients (e.g., the use of picture or visual schedules before and during medical encounters to address communication challenges; use of a shared care plan to facilitate continuity of care or collaboration between HCPs). There is additional work required, however, to develop complete and comprehensive guidelines on how to address particular barriers and what interventions might be most appropriate in what instances as this will be crucial to support organisations in implementing effective practices to improve care. Finally, within studies evaluating patient-focused interventions that were intended to change, or teach, behaviours, participants typically received 1:1 behavioural interventions in their educational setting which yielded uniformly positive outcomes. There is some research (93-95) demonstrating the efficacy of technologies in facilitating the delivery of parent-mediated behavioural intervention to autistic children. Researchers could explore capitalising on technology to allow such interventions to be accessed more readily by autistic individuals and caregivers or provided more commonly through healthcare services or organisations.

The most important measure of the impact of an intervention to improve access to care and care experiences must be whether it actually improves delivery of care, care experiences, and health outcomes for autistic persons. Included studies typically relied on measures of Behaviour (54.8%; Kirkpatrick Level 3) when evaluating intervention outcomes, most commonly the behaviour of the autistic individual undergoing a medical procedure, followed by measures of Reactions (45.2%; Kirkpatrick Level 1), typically of HCPs to an educational intervention. More than 90% of studies using single-subject research designs used measures of behaviour which, again, likely reflects that these studies were typically behaviour analytic in nature and that the measurement of behaviour is a cornerstone of applied behaviour analysis (96). There was greater variability in outcome measures utilised within studies employing groups designs which likely reflects that these studies have emerged from a variety of disciplines and that a variety of constructs (e.g., knowledge, attitudes) were targeted within the interventions. Notably, less than one fifth of studies used measures that were classified as Level 4 measures on Kirkpatrick's evaluation hierarchy (49). Level 4 measures consider 'business results' (97) or impact upon the practice of the organisation and/or improvements for employees or service-users. Given data on increased morbidity and mortality (3-7), unmet healthcare needs (8), and poorer care experiences (8, 9) in autism, we argue that future studies evaluating interventions must assess for stronger indicators of change and impact (e.g., measures of health of autistic persons, measures of patient comfort and/or distress during HCP consultations). It is crucial also that studies measure what matters to autistic patients (98). Research which engages the autistic community to determine preferred outcome measures, and/or which considers what routinely collected healthcare data may offer useful insights, would be of much use in advancing knowledge on how to most efficiently and effectively determine the effect an intervention has had for patients. The measurement of intervention outcomes at the higher Kirkpatrick levels is essential for engaging policymakers and securing financial support (99). Stronger data to support intervention effectiveness may also encourage adoption or uptake of interventions thereby 'spreading' positive change in the physical healthcare of autistic individuals.

Limitations

This review had a number of limitations. First, we excluded studies that delivered interventions focused solely on improving screening and/or diagnosis of autism by healthcare professionals and within included studies any related data were not extracted. We recognise that the identification of a patient as autistic may result in better accommodation of the patient's needs and improved healthcare provision. However, these studies are well-reviewed elsewhere (40, 100) and were not considered to have the specific focus on improving physical healthcare access or care experiences required.

Second, the focus on interventions to improve physical healthcare access and/or experiences only should be noted. Autistic individuals are at a greater risk for experiencing psychiatric conditions (3) so quality of care in mental healthcare services is important. Similarly, autistic individuals may be more likely to experience poor dental or oral health (e.g., caries, receding gums; (101, 102). However, as the barriers to physical healthcare access have been delineated (22, 24), and may differ from the barriers experiences within other types of services, it was considered most appropriate to focus on interventions that were implemented and evaluated in such settings. Future research that synthesises data on the barriers to accessing mental, dental, or other forms of healthcare services for autistic individuals, and interventions to address these issues, is recommended.

Finally, the decision to include grey literature within a review can be contentious. There is a lack of established best practice in how to search and engage with grey literature, searches of the grey literature may not be replicable by others, the return on resources invested for identification of grey literature can be limited, and issues with the interpretation or extraction of data within grey literature can arise due to poor methodological quality and/or poor reporting (103-106). There is some empirical evidence to suggest that the methodological quality of grey literature studies is poorer than that of peer-reviewed studies (104, 107). This is an important finding as it suggests the inclusion of grey literature could constitute a threat to the internal validity of a systematic review as it may lack the methodology and controls to address the research question. These are issues of which a reader should be aware. However, prestigious evidence-synthesis organisations such as Cochrane (108) and the Campbell Collaboration (109) do recommend searches of the grey literature. Within systematic reviews focused on interventions, and their effectiveness, the non-inclusion of grey literature can lead to a problematic overestimation of effect size or overly favourable judgments of intervention effects (36). Further, grey literature searches are particularly recommended where a small body of research exists, there is a lack of consensus on appropriate outcome measurement, and where the context of the intervention is important (103). Accordingly, we did consider grey literature for inclusion in the current review. Our search tactics were intended to identify grey literature from English speaking countries through the use of Google (location set to UK; first 100 returns), Google Scholar (location set to UK; first 100 returns), Ethos, and OpenGrey. Specified search locations impacts ordering of returns on Google databases. Ethos and OpenGrey are largely focused on grey literature arising from Europe or the UK. Therefore, it is important to note that our grey literature search tactics may not have adequately identified papers published outside of Europe. In total, one grey literature study (76) met our inclusion criteria and has been integrated within the current review to provide as complete as possible of an overview of work to-date in this area and to showcase the full extent of interventions which have been trialled in this area. This study was reviewed using the QATSDD, and the mean quality score achieved (i.e., 18.3) was similar to many of the peer-reviewed studies (e.g., Mean QATSDD scores for groups designs studies=17.7, range 9-28).

Recommendations for future research

The synthesis conducted allows for a number of recommendations for future research to be offered. First, the knowledge, behaviour, and attitudes of HCP are a recognised barrier to healthcare access for autistic individuals (24). However, a majority of provider-focused interventions consisted of educational interventions only, and in most cases, these were largely lecture-based. However, it is well recognised that the change resulting from education alone is minimal. It is essential that future research on educational interventions requires active engagement by participants and affords attendees relevant opportunities to practice target behaviours/knowledge/attitudes and receive feedback if training is to result in changes in behaviour that generalise to the clinical environment and into patient consultations. The use of simulation is becoming increasingly commonplace in healthcare research and practice (110) as a means of recreating or replicating patients, clinical situations, and/or facilities. Simulation has the potential to allow physicians to engage in targeted behaviours outside of the clinical setting and to receive corrective feedback which may result in improved clinical performance. Indeed, one included study described the use of an autistic standardised patient (i.e., actor portraying a patient with a particular condition) among nursing students which was reported to impact on communication skills, along with students' critical thinking, prioritisation skills, and patient assessment abilities (75). Simulation could be coupled with in vivo coaching and feedback from an expert to teach healthcare workers how to appropriately perform key non-technical skills (e.g., communication, empathy, shared decision making) and technical skills (e.g., physical exams, blood draws, MRI scans) when caring for autistic patients. Other applications of simulation are possible too. For example, the use of virtual patients to improve knowledge of autism or the use of high-fidelity simulations to explore healthcare workers' implementation of accommodations as outlined in personalised care plans for autistic patients. Future research which further considers the potential for simulation to yield more effective educational interventions is therefore recommended. However, education or training is just one means of improving healthcare delivery and services for autistic patients. Researchers seeking to develop, implement and evaluate provider-focused interventions should therefore also consider how other forms of provider-focused interventions such as individualised care plans, checklists, guidelines, and technological supports may serve to more effectively improve the experience of autistic patients.

Second, studies scored poorly in the quality assessment in relation to user involvement in the design of the research. Only three studies (59, 77, 83) indicated any involvement of autistic persons or their families in the development of the research or intervention that was reported upon. The rationale for including autistic individuals in the prioritisation, planning, and conduct of research relating to autism has been explicated and well-argued (111, 112). Although such partnerships may encounter challenges (113), such involvement will yield data that are more socially valid and may result in improved translation of research and better outcomes for autistic people (111). Social validity relates to the importance of intervention targets (i.e., is this an appropriate or useful goal?), the appropriateness of the intervention procedures (i.e., are the intervention procedure acceptable?), and the perceived importance of the outcomes observed (i.e., are stakeholders pleased with the results of the intervention? 114) and should be assessed via engagement with stakeholders (i.e., the individual participating within the interventions and those close to them). Social validity has been long discussed in relation to behavioural interventions, but its measurement or consideration has not been optimal (114, 115). It is essential that the design of future research, and interventions in this area, is inclusive of autistic individuals, and the caregivers or family members of autistic individuals as appropriate. This will contribute to ensuring that interventions are maximally socially valid and may contribute to clarifying what interventions should be prioritised.

Relatedly, it would be of much use to engage stakeholders (e.g., autistic persons, the caregivers of autistic persons, HCPs, healthcare managers) in a process involving the mapping of barriers to relevant interventions. Researchers have previously attempted to provide suggestions for interventions to address specific barriers to physical healthcare access (e.g., use of picture/visual schedules as a means of addressing communication difficulties, development and delivery of training or education to address issues relating to provider knowledge or skills; 24). However, there is a need to complete this process more systematically, to include a comprehensive overview of existing interventions such as that offered in the current review, and to conduct the process in a way that is inclusive of the stakeholders who will be responsible for the delivery of the intervention or who will experience the implementation of the intervention. Intervention mapping approaches such as this are growing in popularity and have been used with patient populations including patients with cancer, patients experiencing mental health issues, patients with HIV and others (116). Such a process may valuably identify frequent or pervasive barriers that are not adequately addressed by existing or previously-trialled interventions, may lead to suggestions for novel interventions that could be evaluated, and, through the consideration of the frequency with which specific barriers are reported within research, may assist with the prioritisation of interventions for evaluation. There is a clear interest in contributing to the improvement of healthcare services among the autistic community (28) so such an exercise may be well received and yield good engagement.

Next, given the relative dearth of literature focused on interventions to improve physical healthcare access this review has synthesised interventions implemented across all types of healthcare settings. However, it is important to consider differences, or specific challenges or procedures, which may exist across healthcare settings and how these may be addressed. For example, the experience of an autistic patient receiving routine preventative care within a primary care setting from a known family doctor/general practitioner is likely to be quite different to that of an autistic patient admitted to an emergency department while acutely unwell or an autistic patient admitted to hospital for scheduled/elective surgery. The use of patient narratives is a recognised means of informing quality improvement initiatives in healthcare (117, 118) and may be a useful means of furthering understanding of care within specific settings or in relation to specific care services. Alternatively, other qualitative methodologies may usefully elucidate patient experiences. For example, Donovan (119) describes engagement with autistic women via semi-structured interview in order to develop an understanding of their experiences and particular challenges or difficulties encountered. Nicholas et al. (120) employed semi-structured interviews to develop an understanding of the experiences of families with an autistic child required to attend the emergency department. As research on the healthcare experiences of autistic individuals increases, such data may usefully facilitate quality improvement in specific services or specialties.

Finally, included studies typically provided limited detail on the implementation of the intervention. This issue of insufficient description of interventions has been identified previously (121, 122), has been demonstrated to hinder replication of interventions (123) by other researchers, and is suggested to contribute substantially to the ‘waste’ of healthcare research (121) as it precludes the implementation or re-creation of interventions by those working on-the-ground. Further, the effects of interventions which are informed, or delivered, by individuals with high levels of training or expertise, or which are well resourced in terms of personnel or funding, may not be generalisable to other contexts or settings. Therefore, it is essential that future research provides complete detail on intervention content (e.g., components, materials), agent (e.g., expertise/qualifications, supports provided), setting, delivery of intervention (e.g., frequency, schedule), and any flexibility in delivery of interventions (121).

Conclusion

The need to adapt healthcare services for autistic persons is recognised as a priority. However, only a small body of literature internationally has considered how this can be effectively done. Further research is imperative and should look beyond the autistic individual to how provider behaviour and healthcare organisations must alter. The design of future research must be inclusive of the autistic community, must measure what matters, and must offer complete detail on interventions implemented.

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Chapter 7.
General Discussion

Introduction

Improving the physical health and the quality of healthcare for people on the autism spectrum has been repeatedly identified as a research priority by the autism community and their supporters (1, 2). Thus, this thesis examined the barriers to healthcare experienced by people on the autism spectrum and considered how these might be addressed. Five inter-related studies were conducted to help advance understanding of barriers to accessing quality care for people on the autism spectrum from the perspectives of autistic individuals, caregivers, and healthcare providers (HCPs). Although the evidence base for barriers to healthcare access for the autism community has grown over the previous decade, there is still very little literature in this area specific to Ireland. The research presented herein was designed to address this gap with the aim of providing guidance for future intervention development and quality improvement initiatives. This final chapter of the thesis will consider the research findings in the context of existing literature and discuss the implications for future research, policy, and practice.

Placing the research findings in context of existing literature

The findings from the five studies reported in this thesis demonstrated that barriers to healthcare experienced by autistic individuals are prevalent and occur at the level of the patient, HCP, and system. The findings also demonstrate that these barriers can have substantial impacts on the health and healthcare experiences of autistic individuals. Finally, the findings indicate that a small body of research has formally evaluated interventions to improve healthcare experiences for autistic individuals but that more efforts are needed to ensure that such interventions are acceptable, feasible and target what matters to the relevant stakeholders. This section will discuss these main findings of the research in the context of the existing literature on healthcare access for the autism community and existing theories of access to healthcare.

Barriers occur across the healthcare system

A consistent finding across all five studies included in this thesis was that barriers occur at the levels of the patient, HCP, and system. This finding is consistent with previous research (e.g., 3, 4). However, the current thesis adds to the existing literature by delineating the barriers experienced, demonstrating that barriers exist regardless of age and the presence/absence of co-occurring intellectual disability (ID), and indicating that there may be differences in the barriers perceived by various stakeholders. The taxonomy resulting from the systematic review described in Chapter 1 outlined the various categories of barriers. While all of these barriers are important, the following paragraphs will discuss the barriers which were most commonly reported across the studies in this thesis, as well as the wider literature. As these barriers appear to be among the most severe and/or impactful on the accessibility of healthcare for autistic individuals, it is recommended that these should perhaps be the initial focus for interventions.

Barriers associated with autism-related characteristics

Similar to previous research (3, 5-8), patient-level barriers were commonly reported by all three stakeholder groups (autistic individuals, caregivers, and HCPs) across the studies in the current thesis. Although there was variability in which patient-level barriers were reported by different stakeholders, communication and social issues were consistently reported as an important barrier by all three groups. In Chapter 2 (9), communication and social issues were among the most common barriers reported by all three groups. In Chapter 3 (10), the autistic individual's difficulties communicating their thoughts, wants or needs was endorsed by 53% of caregivers as occurring 'often' or 'very often', while 36% of caregivers endorsed this as having a severe impact on care. Additionally, the autistic individual's difficulty identifying, and reporting pain/symptoms was endorsed as occurring 'often' or 'very often' by 62% of caregivers, while 40% of caregivers endorsed this as a severe barrier. Similarly, communication difficulties were endorsed as occurring 'often' or 'very often' by 73% of physicians in Chapter 4; While in Chapter 5, communication or social issues were identified in 87% of the described healthcare encounters. These findings are unsurprising as communication and social atypicalities are core to the diagnostic criteria for autism (11) and have been consistently associated with challenges in healthcare regardless of age or the presence of co-occurring ID (8, 12-15).

Barriers related to communication have also been found to interact with other barriers. For example, the systematic review in Chapter 2 (9) demonstrated that healthcare settings can cause anxiety which can impact on communication abilities, even for those whom communication does not typically pose a substantial problem (13, 14). Thus, it is important that HCPs are aware of the variability in the communication needs of autistic individuals (3, 5, 16). This suggests that communication-related barriers may be best addressed by improving HCPs' ability to identify and implement adjustments to care that facilitate better communication with autistic individuals, such as allowing extra time during consultations (6, 16).

A second patient-level barrier that appeared consistently throughout this thesis, and in the wider literature, are difficulties with waiting and waiting areas (14, 17-19). Difficulties with waiting are particularly problematic as they appear to interact with, or exacerbate, other common patient-related issues. For example, waiting areas can cause anxiety and sensory overload due to the lights, sounds and smells, the unfamiliarity of the people and environment, the disruption to routine, unwanted social contact, and unpredictable waiting times (3, 6, 14, 19). Thus, interventions and adjustments aimed at improving the waiting experience are recommended (16). Such adjustments can be simple, such as allowing the person to wait outside until their appointment (14), or giving the autistic individual an appointment time that is likely to be on schedule (e.g., first appointment in the morning/after lunch (7, 20); or can be more resource intensive such as installing a sensory room in which the autistic individual can wait (21, 22).

HCP-related barriers

All three stakeholder groups commonly reported HCP-level barriers, although, similar to previous research, these were more commonly reported by autistic individuals and caregivers than HCPs (4, 17, 23-26). At the HCP-level, a lack of HCP knowledge about autism and caring for autistic individuals is a key barrier to healthcare access that is consistently reported in the literature by autistic individuals, caregivers, and HCPs (24, 27-33) and this is supported by the findings from the studies in the current thesis. In Chapter 2 (9), a lack of HCP knowledge was commonly reported by caregivers and HCPs, but interestingly was only identified in one study which involved autistic participants. Though it must be borne in mind that there were comparatively fewer studies which considered the perspectives of autistic participants. In Chapter 3(10), 53% of caregivers endorsed a lack of HCP knowledge about autism as occurring ‘often’ or ‘very often’, and 33% indicated that this was a severe barrier. In Chapter 4, 63% of physicians indicated that this barrier occurred ‘often’ or ‘very often’. While in Chapter 5, a lack of physician knowledge about autism was reported in 48% of the described encounters. These findings indicate that there is a pressing need to improve autism education or training for HCPs and that any such training needs to move beyond categorical knowledge of autism, to identifying and implementing appropriate reasonable adjustments for individual and often heterogeneous autistic patients (6, 28). This is important because a number of studies have shown that even when autism knowledge is high among HCPs, confidence in providing care tends to remain low (28, 29, 34; Chapter 4).

It is important to not focus solely on autism training for HCPs to address this barrier, however. There is a myriad of research indicating that HCPs require training in several complex areas such as ADHD (35), suicide (36), and dementia (37), thus, it is not practical or cost effective to expect HCPs to undergo training to address every clinical area in which they lack full knowledge. Further, too much focus on training may detract from the systemic factors that impact on HCPs’ abilities to provide quality healthcare such as time constraints, a lack of resources and financial constraints (6, 28, 38-40). Thus, efforts are needed to examine other strategies to help HCPs to treat autistic patients such as embedding toolkits into existing technologies or resources, developing and implementing individualised care plans, the use of checklists and guidelines, and allocating more resources that would allow for adjustments such as longer consultation times as necessary (3, 16, 34, 41)

A second HCP-related barrier that featured prominently across the studies in this thesis, and in the existing literature, related to the HCP ignoring or not listening to patient or caregiver concerns and/or expertise about autism (4, 8, 17, 24, 42). Feeling ignored or not listened to has been shown to have a negative impact on patient satisfaction with the quality of care received and patient-provider relationships (42-44), which in turn have been shown to impact on treatment adherence (45) and health (46, 47). Various tools exist to facilitate more effective patient-provider communication. For example, hospital passports have been endorsed by the autism community and have demonstrated effectiveness at improving patient-provider communication for individuals with IDD in emergency departments (48, 49). However, research has indicated that HCPs do not consistently engage with such tools (40, 41). Further work is needed, therefore, to establish the best ways of raising awareness, supporting implementation, and ensuring consistent use of available communication resources in healthcare facilities.

Healthcare system-related barriers

As found by other researchers, system level barriers were more commonly reported by HCPs as compared to caregivers and autistic individuals (50, 51-53). Previous research has suggested that patients are more likely to see more proximal factors such as processes of care and interpersonal dynamics of care (51). Nevertheless, some system-level barriers were identified by caregivers as well as HCPs. A lack of coordination between HCPs and services is one of the most consistently reported system-related barriers in the literature, especially by caregivers and HCPs, and this was supported by the studies in the current thesis (3, 5, 33, 54). In Chapter 2 (9), a lack of continuity or collaboration between services and HCPs was the most commonly reported barrier across the included studies. In Chapter 3 (10), difficulties following up on the autistic person's care was endorsed as occurring 'often' or 'very often' by 23% of caregivers and this was endorsed as severe by 6% of caregivers. In Chapter 4, 70% of physicians endorsed a lack of coordination between services as occurring 'often' or 'very often'. While in chapter 5, this barrier was reported in 11% (*n*=5) of the described encounters.

Care coordination is particularly important for autistic patients due to the multitude of co-occurring conditions often experienced that necessitate care across a variety of services, specialties, and HCPs (33, 44). GPs in particular have consistently reported issues around identifying appropriate referral pathways and securing referrals to mental health or other specialty services for people on the autism spectrum (28, 55). Indeed, similar to previous research, many of the GPs who participated in Study 5 (Chapter 6; 56) of this thesis expressed frustration related to obtaining mental healthcare appointments for their autistic patients and reported being bounced around between services (28, 57). This consistent citing of poor care coordination emphasises the need for system/organisation-level interventions to establish reliable channels or pathways between services required by autistic individuals (55). Electronic health records may offer one means of effectively transferring information between services and thus warrant investigation in relation to the care of autistic individuals (34, 42, 58-60). When attempting to improve referrals and coordination between services it is important to ensure that clear lines of responsibility are established, procedures are standardised, all HCPs and services involved possess a shared mental model of what the coordination process involves, and that there are adequate resources for patient transition and follow-up, as failing to do so has been shown to hamper effective coordination (59).

A number of studies have recommended autism-specific case coordinators who can facilitate communication between all relevant parties (HCPs, patients, caregivers), provide case management, assist with healthcare navigation and referrals and/or provide information and support to empower families to coordinate care (43, 61, 62). Such interventions would require additional resources; however, this is problematic as a lack of resources is a second common system-level barrier often reported by HCPs for providing adequate care to their autistic patients (25, 28, 33, 39, 43, 55, 63). Commonly cited resource constraints include a lack of time, a lack of financial reimbursement and a lack of support for HCPs (28, 43, 63-65). A lack of time during consultations is also commonly reported by caregivers and autistic adults (6, 42). This has specific implications for autistic individuals due to the extra time that is often required to process information and formulate responses and questions (6, 12, 66). HCPs often report that, although they are aware of the need for longer appointment times, they do not have the resources or the support to accommodate this (28, 63). Reasons cited in the current thesis and in the existing literature include a lack of support from management, financial disincentives due to a lack of reimbursement and heavy caseloads (63, 67; Chapter 5). Thus, consideration is needed on how to facilitate longer consultation times for autistic patients, without negatively impacting on the HCP's workload or financial reimbursement (33). In terms of other resources, several studies have highlighted that HCPs indicate a need for more staff and administrative support in order to support case management and care coordination for autistic patients (25). Improving resource availability will require additional funding and investment, however, without such commitments through governmental policy, many of the barriers faced by autistic individuals and HCPs in healthcare are unlikely to be solved in the long term (33, 68).

Barriers impact on healthcare and health

The findings from this thesis indicate that experiencing barriers to care can negatively impact autistic individuals' experiences of healthcare and their physical health. Over a quarter (*n*=12; 26%) of the encounters described in Chapter 5 (Study 4) were rated as high severity (i.e., having a severe impact on the patient). Of these, 58% (*n*=7) related to staff ignoring severe patient distress which often involved restraining the patient against their will to complete a medical procedure. The remaining four encounters that were rated as 'high severity' related to incorrect medications being prescribed, being unable to access specialist care, being refused a healthcare appointment, and a missed medical diagnosis. Examination of the barriers that were reported during the encounters where staff ignored severe patient distress revealed that the most commonly occurring barriers were all HCP-related and included a lack of HCP knowledge about autism, poor HCP communication, HCPs ignoring patient/caregiver concerns and expertise, stigma or negative perceptions, and HCP inflexibility. These findings are not surprising as previous literature has reported similar barriers occurring during distressing or substandard experiences in healthcare (13, 24, 69). Lum et al. (13), for example, found that autistic women reported receiving less support and information during childbirth compared to non-autistic controls; while caregivers in another study described the perioperative care process as ‘a trying and disgraceful scenario’ and ‘a hopeless struggle’ which encompassed ‘unspeakable suffering’ (69). Although the literature indicates that HCPs are often aware of barriers such as inadequate autism knowledge and communication difficulties, they are less likely to report challenges associated with their own communication style and flexibility when providing care, both of which are commonly reported by autistic individuals and caregivers (4, 12, 13, 24, 62, 66). Thus, HCPs may need support to reflect on how to adapt their own behaviour to facilitate better consultations and improve care experiences.

System-related barriers also impact on the care experience; for example, Nicholas et al. (26) found that in addition to HCP-level barriers (e.g., not including parents in care, a lack of autism knowledge), the process of care in the ED and time-related issues also contributed to negative care experiences for autistic children, whereby children were not given enough time to acclimate to the prospect of a medical procedure, and sedation and restraints were used to perform procedures as quickly as possible. Similar findings have been observed elsewhere, in various healthcare settings including non-emergency hospital settings and primary care (14, 15, 17, 24, 62, 69). Negative healthcare experiences generally result in poor satisfaction with care (26, 70-72). Thus, it is necessary to improve care experiences as poor satisfaction with care is associated with various adverse outcomes including poor adherence to treatment (73, 74), poor engagement with preventative healthcare (75, 76), increased depressive and stress symptoms in caregivers (77), and poor health outcomes (78). Since autistic individuals have been found to report poorer healthcare experiences and satisfaction with care than their peers in the general population (72), they may be more at risk of experiencing these related adverse outcomes further highlighting the health inequities experienced by the autistic population.

In addition to impacting healthcare experiences, barriers can have a negative impact on health. The described encounters in Chapter 5 resulted in a range of adverse outcomes including delayed or missed diagnoses and procedures, incorrect medications being prescribed and being unable to access specialist care. This echoes findings from a recent study conducted in the UK and Ireland in which difficulties attending healthcare appointments were correlated with untreated physical and mental health conditions, not attending specialist referrals or screening programmes and requiring extensive treatment or surgery due to late presentations and untreated potentially life threatening conditions (7). In addition, research has shown that autistic individuals experience lower rates of preventative care, including vaccinations (71, 72, 79), well-child visits (79), gynaecological visits (80), and cancer screening (72, 80).

Access to preventative care is essential as research suggests it can improve the early detection of new health conditions, reduce hospital admissions, and reduce premature mortality (81-87). Thus, urgent attention is required to reduce unmet preventative care needs for the autistic population. In Chapter 3 (10), significant correlations were observed between experiencing more unmet healthcare needs and the frequency of barriers endorsed by caregivers. In particular having unmet needs was associated with more frequent HCP and system-level barriers. This is also consistent with previous research. For example, Karpur et al. (88) found that unmet healthcare needs for autistic children were associated with system-related factors such as health insurance coverage and access to family centred care, while Farmer et al. (89) found that access to a medical home (i.e., person-centred, coordinated care) was associated with fewer unmet healthcare needs. Access to person-centred coordinated care has been suggested to improve health outcomes for children with special healthcare needs (90, 91). Unfortunately, research indicates that autistic individuals are less likely than individuals with other special healthcare needs to receive a person-centred, coordinated style of care (44, 92). As outlined in Chapter 1, experiencing unmet healthcare needs can translate into poor health outcomes and higher mortality rates for autistic individuals. Thus, more efforts are required to facilitate access to person-centred care and reduce unmet needs and improve care delivery for the autistic community.

Theoretical considerations

As discussed in Chapter 1, access is a complex concept that has been conceptualised and defined in various ways in previous decades. However, although these theories vary in their specific definitions and components, the majority place at least some focus on the importance of the ‘fit’ or interaction between the services and the population (e.g., 93-95). The studies in this thesis were not designed to test any one specific theory and as a result there has been no validating, refuting, or improving any one theory. The findings from this thesis do, however, support the idea that access to healthcare for autistic individuals is an interaction between the supply- and demand-side characteristics as both were identified as barriers in Studies 1-4 (Chapters 2-5). On the supply-side are HCP-related barriers (e.g., autism knowledge, HCP inflexibility, poor HCP communication) and system-related barriers (e.g., lack of coordination between services, time/resource constraints, location issues). On the demand-side are patient-related barriers (e.g., communication/social issues, sensory issues, complexity of family involvement). Thus, access is influenced by various factors which correspond to the different dimensions in a number of the access theories. However, based on the findings of this thesis, the willingness of services to adapt to the needs of the consumer, which is clearly an important factor influencing the accessibility of healthcare for autistic individuals (12, 41) is either missing from existing theories or not emphasised strongly enough. *Accommodation* is a dimension in Penchansky & Thomas’s theory (93) and Levesque et al.’s theory (94). However, the definition of this dimension in these theories relates to how services are organised to accept patients (e.g., opening hours, whether patients can walk in without appointments etc.). There is no mention of the adaptability of services to suit the unique needs of individual patients which is required by autistic individuals. *Acceptability* is another dimension which is likely important for autistic individuals (93, 96, 97). However, this tends to relate to the social and/or cultural factors (i.e., the attitudes of patients towards characteristics of the services providers and vice versa). Thus, it is likely that further conceptual work is needed to expand on some of these theories of access in order to make them fully applicable to the autism community. Such expansion should include specific references to making reasonable adjustments to care to suit the needs of individual patients.

Of the existing theories, Lévesque et al.’s (94) is the one that is perhaps most applicable to the research conducted in this thesis. This theory takes into account specific demand-side characteristics such as health literacy and the patient’s knowledge about healthcare options and their individual rights, which have been implicated in relation to access to care for autistic individuals (6, 8, 12). Richard et al. (98) used Levesque’s theory to collectively assess initiatives used to improve access to care for various vulnerable populations (e.g., people with disabilities, refugees, people experiencing homelessness, LBGTI+ community) from an equity perspective. Richard et al.’s (98) findings indicated that most initiatives targeted supply-side characteristics, with a very small percentage targeting demand-side characteristics and just over a quarter targeting both supply and demand-side characteristics. The findings from Study 5 (Chapter 6; 56) in this thesis may contradict the findings from Richard et al. (98) as Chapter 6 (56) found that interventions predominantly targeted demand-side characteristics (i.e., patient-related barriers such as the individual’s ability to tolerate healthcare procedures). Research on improving access for individuals with chronic conditions suggests that interventions should be more patient-oriented, focused on self-management and health literacy (99). However, the findings from this thesis suggest that for autism, there needs to be more focus on the system and HCP as these have received substantially less attention than interventions focused on changing the individual (Chapter 6; 56). This finding is similar to research carried out with people with ID. Although there has been some consideration of HCP and system-level interventions for people with ID (100, 101), there has been more of a tendency to focus on patient-level interventions (100, 102). The discrepancies between the findings of supply-side dominance in the Richard et al.'s (98) study and the findings of demand-side dominance outlined in the Chapter 6 (56) may be due to the fact that the interventions in Richard et al. (98) targeted a wide array of vulnerable populations. This underscores the importance of examining autism as a separate population as there are discrepancies between what is required and appropriate to improve access for autistic people compared to other vulnerable populations (72, 103).

Numerous previous studies have concluded that equity of access to healthcare requires that interventions consider the social and health determinants of the population, the needs of the population as well as the resources available to them (98, 104-106) and this appears to hold true for autistic individuals. However, questions still remain around establishing the optimal combination of supply- and demand-side dimensions of access. As theories of access continue to evolve in relation to the autism community, there is a need to aim for a more person-centred approach to defining access with equal emphasis on both supply- and demand-side characteristics, similar to that of Lévesque et al. (94). This is likely imperative to gain a full picture of access for the autism community and would help to identify gaps which could guide interventions to target specific dimensions when attempting to improve access. Indeed, it has been recommended that interventions aimed at improving practice in healthcare should be based on mapping pre-identified determinants, including barriers and enablers of healthcare with theoretical constructs (107). Incorporating theory into intervention development in this way is important because theory can guide the development, implementation and evaluation of an intervention and provide insight into how an intervention works (108, 109). Further, interventions based on theory have been shown to improve professional practice and are more likely to be adopted and implemented in practice (110).

Considerations for intervention development

Although the barriers to healthcare access have been well considered in this thesis and elsewhere (3, 5, 6, 12) there is less guidance on how to improve services for autistic individuals. This must be the next step in improving access to healthcare for autistic individuals. Some progression has been made in relation to improving health and healthcare for individuals with ID such as systematically implementing health checks (111). However, although a growing body of research has begun to highlight suggestions for how to improve care for autistic individuals specifically (16, 103, 112, 113). Until recently there had been no attempt to consider the collective empirical evidence supporting the effectiveness of these recommendations or their outcomes. Chapter 6 (56) reviewed the small body of research that has formally evaluated interventions. The findings indicated that, to date, interventions have predominantly focused on changing the patient’s behaviour (e.g., teaching the autistic individuals to cooperate with injections; 114) with less focus on changing HCP behaviour (e.g., increasing HCP knowledge about autism; 115) or the system/organisation (e.g., modifying the physical environment; 116). However, as the studies in the current thesis and previous research have indicated, autistic individuals and caregivers commonly report experiencing substantial HCP-level barriers to care such as a lack of autism knowledge among HCPs, and a lack of HCP flexibility for implementing adjustments to care (3, 5, 7, 8, 12, 54). Further, HCPs have reported system-level barriers including a lack of resources and poor coordination between services, to providing quality care to their autistic patients (28, 39, 63, 67). Thus, it is essential that future intervention development focuses on targeting HCP- and system-level barriers as this is a clear gap in the extant body of literature. However, it must be noted that research has consistently shown that system-level interventions are associated with substantial challenges including inefficient systems of care (e.g., difficulties with cross-system communication, dysfunctional medical record systems), difficulties engaging patients and families, HCP attitudes or scepticism, staffing and resource issues, a lack of organisational support, complexity of interventions, and organisational cultures (117-119). Thus, system-level interventions are likely be more complex and resource intensive, but in order to effect meaningful, lasting change to the healthcare and health of autistic individuals, such interventions are necessary.

In addition, it is also necessary to establish an evidence base pertaining to the various sub-populations within the autistic community. As outlined in Chapter 1, the autism spectrum is very heterogenous, thus, a one-size-fits all approach to improving care is unlikely to be effective (120). It is, therefore, recommended that future work investigate the effectiveness of interventions for the various sub-populations of the autism community, for example, those who have co-occurring ID or those who experience mental health difficulties. It is also essential to investigate the barriers and potential interventions relevant to autistic individuals from marginalised groups such as ethnic minorities or the LGBTQI+ community, as members of these communities may be at even greater risk of experiencing health inequities and may face very specific barriers to care which require more nuanced interventions (121, 122). Consideration also needs to be given to interventions suitable for different age groups. Chapter 6 (56) highlighted that to date, interventions have focused predominantly on autistic children, with just three interventions involving autistic adults. Although some barriers are likely to overlap (e.g., communication difficulties, sensory difficulties; 6, 12, 26) it is likely that autistic children and adults will have different experiences regarding barriers to care and thus require tailored interventions. For example, adults are less likely to have a caregiver accompanying them to healthcare appointments, and while this may be preferable for some autistic adults (16), it may create challenges for others (e.g., for those who experience slower information processing speeds; 3, 5, 12). The requirement for a supporter may also vary by healthcare setting. For example, an autistic adult may feel comfortable going to their GP alone but may prefer to have a supporter accompany them to the hospital due to the busy unfamiliar environment (16). Thus, in addition to considering interventions for various subgroups of the autistic population, it is also necessary to consider interventions aimed at different healthcare domains, as different settings present different barriers.

It is also necessary to consider how interventions are evaluated as this has implications for appraising the effectiveness of interventions (123). The findings from Chapter 6 (56) indicated that although the outcomes of the reviewed interventions were predominately rated as positive, the majority of these interventions were assessed through measures of behaviour (e.g., the autistic individual’s compliance with a procedure) and participants’ reactions (e.g., HCP satisfaction ratings of educational interventions). More valuable measures of the impact of intervention are whether it actually improves the delivery of care, care experiences, and health outcomes for autistic individuals. Thus, future intervention evaluation should measure what matters to autistic individuals as well as focus on stronger indicators of change and impact by assessing outcomes of the higher levels of Kirkpatrick’s Evaluation Hierarchy, including measuring the impact of the intervention on the organisation, and the employees or service-users (123, 124). Suggestions for how effective future interventions may be developed will be discussed in the recommendations section below.

Recommendations for future research, policy, and practice

Throughout this chapter, recommendations for research, policy, and practice have been highlighted where appropriate. This next section will consider some additional points in more detail.

Recommendations for Research

In addition to the suggestions for future research that have already been outlined in this chapter, there are a number of additional avenues through which future research could build on this thesis, that would greatly contribute to our understanding of barriers to, and inequities in, healthcare for autistic individuals, some of which will be highlighted in this section.

Involvement of autistic persons in intervention development

Future research should comprise participatory research with autistic individuals and their supporters, ensuring that autistic people are involved throughout the entire research process in accordance with current best practice (1, 125). This is especially true for intervention development. Specifically, future research should investigate what interventions the autistic community believe are needed within healthcare. In the current thesis, I have gathered evidence regarding interventions based on the literature (Chapter 6; 56) but only three of these interventions demonstrated any evidence of user involvement in the design (41, 116, 126). Thus, there has been very little input from autistic individuals when designing interventions to improve their healthcare experiences. Interestingly, those interventions which demonstrated user involvement in the design all comprised HCP- or system-focused interventions. The lack of user involvement in research to-date is potentially one reason for the over-reliance on interventions targeting the individual as opposed to implementing HCP- and system-level changes. It is also important to consult HCPs regarding intervention development to ensure that interventions target barriers to providing care that are important to HCPs (33, 65, 67). Thus, co-designing interventions together with all stakeholders would help to ensure that any planned initiatives are feasible, acceptable, and targeting the things that matter to the relevant stakeholders (125, 127). There now exists useful guidance on conducing participatory research with autistic individuals which should be consulted by researchers going forward in order to effectively involve diverse groups of people in participatory and co-design processes (127-129).

Intervention mapping

Although this thesis did not result in the development of an intervention to improve access to care for autistic individuals, my research has offered the first step towards development of an intervention or interventions to improve care delivery for autistic persons. As a next step, it is recommended that intervention mapping (IM) be conducted, drawing on the data on barriers generated through the various studies which comprise this thesis, and on the identified theories of access described in Chapter 1 (130). Bartholomew et al. (131) developed the intervention mapping (IM) approach as a process of applying theory to intervention development, in which the path from problem identification to problem solving is mapped. The IM protocol (132) provides guidance on how to conduct effective IM through six distinct, but iterative steps, each of which, on completion, provides guidance for the next step. Briefly, these steps are: (1) conduct a needs assessment to identify what needs to be changed and for whom; (2) establish objectives (i.e., identify what beliefs/behaviours should be targeted by intervention); (3) select theory-based intervention methods and practical strategies to change the targeted behaviours/outcomes; (4) design and produce programme components and organise these into an structured programme/intervention, and pilot test the intervention; (5) plan for adoption, implementation and sustainability of the intervention in real life contexts; (6) generate an evaluation plan to assess the impact on the intervention. The completion of all six steps produces a blueprint for designing, implementing, and evaluating targeted interventions which are based on theoretical, empirical, and practical information (132, 133). IM has become a popular approach in healthcare research and has been used develop interventions for patients with cancer (134, 135), patients experiencing mental health issues (136, 137), and people living with HIV/AIDS (138, 139), among others (140). In a review of 22 IM studies, Garba & Gadanya (141) found that IM resulted in significant uptake of disease prevention programmes including cervical screening (142), influenza vaccination (143), and mammography (144). Thus, future research should engage the autism community and HCPs in IM to ensure the development of effective interventions. This would also allow interventions identified by the relevant stakeholders to be mapped to the interventions identified in the literature (Chapter 6; 56) to assess whether or not targets and outcomes are aligned. This information could then be used to guide future intervention development and evaluation, taking into account the feasibility, acceptability and cost-effectiveness of interventions.

Understanding the enablers of high-quality care

The current programme of research examined barriers to physical healthcare only. Future research may wish to assess enablers (i.e., factors which contribute to improved care experiences (26) of effective physical healthcare. This information would be particularly useful for informing intervention development and identifying appropriate reasonable adjustments, something with which HCPs consistently report experiencing difficulties (28, 40). As with barriers, it is recommended that standardised tools for assessing enablers are developed. These tools would facilitate direct comparisons to be made across various healthcare settings and support benchmarking across services (145). Brice et al. (16) recently developed a tool for assessing the availability and importance of reasonable adjustments in physical and mental healthcare services. The authors found that although some adjustments were important across both settings (e.g., access to a clinician who understands autism), there were some discrepancies (e.g., a clinician who bases their approach on information provided by the autistic person was deemed more important in relation to mental healthcare services). Thus, although it is likely that some enablers will be important across services (e.g., recognising the patient/caregivers' expertise about autism, ensuring person- or family centred-care and good patient-provider communication; 24, 146, 147), it is also likely that enablers will vary according to service type. For example, what is helpful in a busy ED may not be useful in a quiet primary care office and vice versa. Thus, it is also recommended that assessments are conducted to identify service-specific enablers or reasonable adjustments. Such assessments of enablers could be incorporated in existing barriers tools to offer more comprehensive appraisals of healthcare facilities. A number of systematic reviews have assessed barriers and enablers together for autistic adults with (8) and without (3, 5) co-occurring ID, however, a systematic review focused specifically on facilitators and stratifying this by various sub-populations within the autistic community and healthcare settings, would allow for a more thorough exploration of opportunities to enhance healthcare services for autistic individuals. This may produce more actionable data and could further expand the conceptual framework of access to healthcare for autistic individuals (33, 145).

Other determinants of health equity for autistic individuals

As discussed in Chapter 1, access to healthcare is just one determinant of the substantial health inequities associated with autism. Although, a substantial body of research has indicated that autistic individuals are at a high risk of experiencing many of the other known determinants of health such as social determinants (148), genetic/biological factors (149), and health communication difficulties (8, 13), there has been less focus on the associations between these determinants and the impacts they have on the physical health of autistic individuals. Research has consistently demonstrated that autistic individuals have a high risk of experiencing social determinants of health in particular, including unemployment (150, 151), poverty (152, 153), social disconnectedness (154, 155) and discrimination (156). Although it is well established that unemployment is linked to poor health and wellbeing in the general population (157-159), there is a paucity of research which has formally examined how unemployment impacts on physical health for autistic individuals, despite the high rates of unemployment in this population (150) and the observed links between unemployment and mental health issues for autistic individuals (160). Similarly, it has been well established that social disconnectedness can have adverse impacts on health in the general population (161, 162) and it is known that autistic individuals are at a high risk of experiencing social disconnectedness (163, 164). Yet, little research has examined the links between social disconnectedness and physical health; more research exists examining the impacts of social disconnectedness on mental health for autistic adults (165). Thus, future research is needed to assess the specific associations between the various social determinants and physical health outcomes. Such research would further strengthen the argument for focusing attention on reducing the exposure of autistic individuals to these determinants. In addition to social determinants, health communication and health literacy have also been implicated as an important determinant of health inequities for autistic people (12, 26). However, although research exists which links low levels of health literacy to poor health outcomes (166-168). there is a lack of research examining these links specifically for autistic individuals. Such research is needed so that appropriate interventions can be developed to improve health literacy for people on the autism spectrum. This may involve training for HCPs on effective communication strategies for communicating with autistic patients, or the development of accessible literature in various formats (e.g., audio, electronic, pictorial). Thus, future work is needed to advance knowledge on how exactly each of the known determinants of health inequities impact on physical health for the autistic population in order to guide effective, appropriate interventions to improve health outcomes.

The relationship between barriers and health outcomes

The current thesis has identified that barriers exist, has provided information on the context in which they exist, and the data from the patient narratives have indicated that these barriers can impact on health. Future research could also examine whether certain barriers can be linked to specific health outcomes for autistic persons. These findings support recent research which found that barriers resulted in missed healthcare appointments, with some participants reporting that they had not attended healthcare for serious/life threatening conditions (7, 16). Clearly, barriers can have an impact on health, however, this thesis, has not identified direct links between specific barriers and particular health outcomes. The links between barriers and health outcomes have been examined for other marginalised or disadvantaged groups including ethnic minorities (169) and people in low and middle income countries (170). Establishing links between barriers and health outcomes could be useful for guiding intervention development and the appropriate allocation of resources so as to ensure key barriers are the focus of interventions. In addition to intervention development, the consideration or measurement of health outcomes would also be a valuable means of evaluating interventions. Such evaluations are largely missing from the autism literature to date. As discussed above, Chapter 6 (56) demonstrated that interventions were most often evaluated through assessments of reactions and changes in the autistic person’s behaviour, no intervention measured changes in health status. In order to engage policy makers and secure funding, it is essential that interventions are evaluated at the higher levels of the Kirkpatrick Hierarchy (i.e., objective changes in the organisation or participants; (123, 171). Assessing objective health status is a potential means of assessing changes in participants as a result of intervention and would add substantial value to intervention evaluation. Changes in health status could be useful in cost-benefit analyses of interventions which would guide resource allocation for sustaining successful interventions or discontinuing ineffective interventions (172). Thus, more efforts are needed to develop objective outcome measures such as objective assessments of health status that can be used in research concerning physical healthcare for autistic individuals.

Recommendations for policy and practice

It is key that research serves to influence policy and practice so that research findings have a meaningful impact on the individuals who work in and use the health services. Policy will dictate practice by ensuring adoption of research findings and recommendations into practice at a society-wide level, which is essential for ensuring effective inclusion and supports for autistic individuals.

Need for objective assessments of health status

In addition to monitoring key determinants of health such as access to care, there is also a need to monitor health status as a means of assessing actual equity in health (173). Annual health checks are one useful means of examining objective health status (83, 174). In the UK, annual health checks have been implemented in primary care for people with ID. The purpose of the check is to assess barriers to accessing care, identify unmet healthcare needs, and improve prescribing and care coordination practices (111, 175).

Systematic review evidence has demonstrated that health checks lead to improved detection of new health problems and increases in uptake of preventative health behaviours (e.g., vaccines, cancer screening; 83, 84, 176). It is, therefore, important to develop and implement health checks specifically for people on the autism spectrum as the autistic population experience different barriers and health conditions to people with learning disabilities (103). Structured health checks would likely benefit both HCPs and autistic individuals. The systematic implementation of health checks could provide HCPs with the resources they need to provide personalised care to their autistic patients (e.g., access to online care records) and could help to improve HCP knowledge around autism through resources and training made available through the programme as is the case with the established health checks for people with ID (e.g., best practice guidance documents; e-learning resources; 177). For autistic individuals, implementing health checks could provide a more definite route for accessing services, which is important as autistic individuals and caregivers have noted that navigating the healthcare system can be challenging (12, 54). Finally, the implementation of structured health checks may improve access to, and the quality of, primary care services, which may reduce the over-use of emergency and other tertiary services for autistic people (85-87). This is particularly relevant as research has indicated that the high rates of ED use among autistic individuals are, at least somewhat, accounted for by unmet needs in, or poor access to, primary care (85, 178). Autistica UK and a research group in Newcastle University have begun to develop and trial health checks for autistic individuals (179, 180). No such initiatives exist within the Irish healthcare system, however, and so warrant investigation within an Irish healthcare context. It is recommended that any such investigations should also include a cost-effectiveness analysis to establish, for example, whether the costs of implementing health checks is offset by reduced hospital admissions (87).

Need for improved education and training for healthcare providers

The knowledge, behaviour, and attitudes of HCPs regarding autism are a recognised barrier to healthcare access for the autism community (3, 5, 9, 10, 33) - a finding that is supported by all five studies in this thesis. Thus, a clear implication for future practice is the need for improved education and training for healthcare professionals to enable them to provide high quality care to their autistic patients (28, 33). In the systematic review of interventions (Chapter 6; 56) the findings indicated that the majority of HCP-focused interventions consisted of educational interventions only and these interventions were largely lecture based. Education alone is recognised as having only minimal effects, however (181, 182). It is important therefore that future educational interventions incorporate active engagement of participants in order to allow learners to practice target skills/knowledge/behaviours and receive feedback; this should facilitate generalisation of learned content to the clinical environment (181). Communication skills training is potentially one of the most pressing areas in which HCPs need training as many HCPs self-report difficulties communicating with autistic patients (28, 67). Thus, HCPs should be given more exposure to real-life and case study experiences with autistic individuals as this has been shown to improve HCP communication skills and attitudes (183-188).

The use of simulation in medical education has become common and is an effective means of allowing learners to practice target skills (189). A small body of research has begun to investigate the use of autistic standardised patients for teaching communication skills, and patient assessment abilities in nursing research and has demonstrated encouraging results (190). An important limitation in McIntosh et al.’s (190) study, however, is that the actor was not actually autistic. Research has demonstrated that engaging people who have lived experiences of IDD as simulated patients for training medical trainees is both feasible and recommended in order to produce truly authentic scenarios and experiences for the learner (183, 191-194). Video-based patient narratives can also be used for training purposes and have also demonstrated positive outcomes. Coret and colleagues (183) found that medical students' self-reported ratings of comfort, confidence, and competence for caring for people IDD increased following exposure to video-based patient narratives of people with IDD and encounters with simulated patients who had lived experience of IDD. In addition, the intervention group (i.e., exposure to patient narratives, followed by reflective discussions) also demonstrated higher mean scores on objective assessments of their communication skills during the simulated patient encounters with patients with IDD compared to the control groups who just watched an introductory video lecture about IDD. Thus, future research and training should work to engage autistic individuals as standardised patients in the development and delivery of simulated patient encounters and the recording of patient narratives which could be used as training material. Such investigations should also include more objective assessments of the longer-term impact of such training ventures, including whether the knowledge and skills gained translate to the clinical environment, contribute towards lasting shifts in the perspectives and practices of HCPs, and translate into better healthcare experiences and health outcomes for the autism community. The small body of work engaging people with lived experiences of IDD indicate that there is scope and benefit for implementing such training in undergraduate and postgraduate curricula (183, 191, 192). Thus, educators should work towards implementing such training into all HCP curricula as standard.

Practices must measure barriers and autism-friendliness

This thesis has demonstrated that it is feasible to measure barriers that impede quality healthcare for autistic individuals and that this can be done using a variety of methods. The development of ‘autism-friendly services’ has been highlighted as a research priority by the autism community and their supporters (1). Therefore, healthcare facilities should make efforts to conduct evaluations of their services to determine how ‘autism-friendly’ they are. HCPs could ask their autistic patients to complete the self-report barriers tool developed by Raymaker et al. (12) or the Autism Healthcare Accommodations Tool (AHAT) which is available online to help HCPs to identify what adjustments they need to provide (41, 195). HCPs could also administer the tool developed in Chapter 3 (10) of this thesis to caregivers. This could facilitate conversations between HCPs, patients and/or caregivers about what accommodations are required and, in doing so, facilitate the provision of person-centred care (196). Further, HCPs could complete the tool developed in Chapter 4 of this thesis to enable them to reflect on the barriers they experience within their own settings. In addition, tools such as the Green Light Toolkit, which is available online for assessing the accessibility of mental healthcare services, could be adapted for physical healthcare services (197). The uptake of conducting such assessments could be mandated by policy by incorporating them into any National Autism Plans at government level as has been done in the updated UK Autism Strategy: Fulfilling and Rewarding Lives (198 p.14) The current thesis has also demonstrated the value of collecting qualitative feedback. Patient narratives are an established method of obtaining feedback in healthcare and evidence suggests that patient narratives contain actionable data which can be used to identify strategies for improving care (199-201). Thus, future research should investigate how clinicians could gather and use patient narratives within organisations or practices as feedback for improving their services. It is also recommended that service evaluations use both surveys and patient narratives where possible as it has been demonstrated by the current thesis that the triangulated data generated by both modalities provides much richer insights. Surveys are useful for identifying what barriers exist, but the patient narratives provide information on where, when, why and how barriers occur and illustrate the impact that barriers can have on the patient (202).

Facilities must start systematically and consistently implementing adjustments

In many countries, including the UK and Ireland, there are legal requirements that public services make reasonable adjustments (i.e., doing things differently to normal to ensure that persons with disabilities are not disadvantaged) to improve accessibility of public services for persons with disabilities (203, 204). In addition, various autism-specific guidelines exist which recommend that staff and facilities implement reasonable adjustments for autistic patients (205-209). Further, the need for reasonable adjustments to healthcare has been strongly endorsed by autistic individuals. However, the autism community also report substantial difficulties obtaining the required adjustments, even in the UK where autism-specific legislation exists (6, 7, 16, 210).

Implementing reasonable adjustments is paramount to improving the accessibility of care. Recent research has found that numerous studies have examined the adjustments to care that autistic individuals consider important. Suggested adjustments include clinicians who understand autism, easily identifiable and accessible locations and clinicians, opportunities to ask questions after an appointment, locations with low noise levels, options to book appointments online or via text, emailing a description of the problem to the doctor in advance of the appointment, being able to wait in a quiet area or outside until the appointment, or being given the first or last appointment of the day (7, 16). Similar accommodations have been outlined by caregivers and HCP and include having something to occupy the autistic person in the waiting or exam room (e.g., sensory box), scheduling appointments for certain times of the day to reduce waiting times, allowing extra time and explanation when performing vitals and exams and avoiding unnecessary vitals (7, 55). Thus, adjustments to care do not necessarily require extensive resources, instead what is needed is improved staff training, adaptability and flexibility in care provision and attention to the clinical environment (6, 7, 16).

Despite the potential benefits of adjustments, research has shown that they are not systematically implemented across services and tend to depend on individual HCPs’ knowledge and/or willingness to implement them (40, 55). For example, in a recent study of HCP self-efficacy, Nicolaidis et al. (29) found that 75% of surveyed HCPs were not confident in identifying or implementing reasonable adjustments to care for autistic patients. There now exists an abundance of free online resources which can help HCPs identify potential adjustments (e.g., 41, 48, 211) which HCPs should consult in an effort to improve care delivery for their autistic patients. However, the onus should not be on the HCPs alone. Recent research suggests failures to successfully implement reasonable adjustments may be a result of systemic shortfalls, whereby HCPs do not have sufficient resources, or support from management to identify and implement adjustments (16, 28, 40). Thus, in order for facilities to implement accommodations for autistic people, systemic level reforms are required. This will require more resources but without systemic support, adjustments are unlikely to be consistently implemented across healthcare facilities and will continue to depend on individual HCPs.

National Autism Strategy for Ireland

In Ireland, the United Nations Convention on the Rights of People with Disabilities (UNCRPD; 212) was not ratified until 2018. Ireland was the last country in the European Union (EU) to do so. Thus, Ireland may be considered to be some way behind other EU countries and the UK regarding activity on, and commitment to supporting, the rights of persons with disabilities. The ratification of the UNCRPD means that Ireland is now obliged to promote, protect, and ensure the full employment of human rights by persons with disabilities and ensure that they enjoy full equality under the law. In addition, Ireland’s 2005 Disability Act (203) stipulates that public bodies, including healthcare services, must ensure that their buildings and services are accessible to, and inclusive of, persons with disabilities. However, autistic individuals in Ireland consistently report experiencing discrimination and difficulties accessing public services (213). In addition, there is currently no legislation in Ireland that is specific to autism. Multiple reports have highlighted the difficulties faced by autistic people and their families in Ireland, these include access to education (214), access to employment (215), access to diagnostic and interventions services (214), access to mental healthcare services (216-218) and now this thesis has highlighted the difficulties faced in relation to physical health and healthcare. Thus, Ireland urgently needs to develop and enact a National Autism Strategy in line with the European Charter of Rights for Persons with Autism (219). National Autism Plans and strategies that are implemented at government level are essential for ensuring that meaningful inclusion, empowerment, and supports are achieved for autistic individuals within wider society (198). Autism strategies have been established in the legislative agendas in other EU countries (e.g., Denmark, Hungry, Malta) and the UK and so it is recommended that Ireland look to these for guidance. In recent years, some efforts have been made to develop autism strategies and legislation in Ireland (e.g., 220, 221), however none of these have been signed into law or translated into meaningful action. Currently work is on-going on a new Autism Empowerment Strategy (222) which appears to be considering a more holistic view of autism than the previous two bills which took a more medicalised view (220, 221). This is promising but, based on the findings of this current programme of research, it is imperative this new strategy include specific references to improving the physical health of, and access to physical healthcare services for, people on the autism spectrum in Ireland. Finally, efforts must ensure that this new strategy is actually signed into law.

Research Strengths and Limitations

Strengths

This programme of research had a number of considerable strengths, some of which relating to the individual studies are discussed in Chapters 2-6. The quality of the research conducted is evidenced by the progression of the thesis chapters through the peer review process - four papers (Chapters 2, 3, 4 and 6; 9, 10) have already been published in academic journals, one paper has (Chapter 6; 56) has been accepted for publication; one other paper (Chapters 4) has been resubmitted after addressing peer reviews, and a final paper (Chapter 5) is currently undergoing peer review for the first time. Although not without its criticisms, successfully progressing through the peer-review process is generally accepted as an indication of study quality (223, 224). This section will discuss some of the broader overall strengths of this thesis.

Multi-method approach

The greatest strength of this thesis is the multi-method approach taken to assessing barriers to healthcare for people on the autism spectrum and beginning the process of considering how healthcare might be improved for autistic individuals. The use of quantitative and qualitative data together can provide more nuanced and useful insights into a problem (225-227). Further, quantitative health services research has been criticised for ignoring the patient's lived experience, while qualitative health services research has been accused of being too subjective and difficult to replicate, thus, incorporating both approaches has the potential to avoid such tendencies and balance the findings (228, 229).The use of a multi-method approach, therefore, has allowed for some triangulation of the data, providing a comprehensive, authentic overview of this complex issue of healthcare access for autistic individuals (226, 230, 231). A variety of rigorous methods were applied and adhered to within each of the individual studies in this thesis, including factor analyses and best practice for conducting systematic reviews (232). Further, the CIT technique is an effective but relatively under-utilised method for assessing what helps or hinders the provision of quality care, patient satisfaction and patient safety within healthcare and has allowed for a thorough exploration and richer understanding of the issues relating to accessing healthcare for people on the autism spectrum (233-237).

Multiple stakeholder perspectives

The inclusion of multiple stakeholder groups, including autistic individuals, caregivers and HCPs as participants has allowed for the triangulation of perspectives (238). This triangulation helps to improve the confidence in the research findings and can help reduce bias (239). Incorporating multiple perspectives has also been recommended to obtain a thorough overview of the barriers to accessing care experienced by autistic individuals and to ensure the development of interventions that target what actually matters to those involved (5). It is also important to examine various perspectives so as to identify potential overlapping or divergent issues which may be particularly useful for informing quality improvement initiatives (5, 6). HCPs may not be aware of important barriers being experienced by patients (e.g., executive function-related issues; 12), while patients and caregivers may not be aware of the barriers being experienced by HCPs (e.g., financial constraints; 65). Thus, omitting the perspectives of one group may result in bias or an incomplete picture of the problem (6, 239). Conversely, identifying factors that overlap as causing substantial difficulties for all stakeholders provides valuable information for developing QI initiatives as it may be most efficient to tackle barriers that are common to all three stakeholder groups (6). The triangulation of perspectives in the current thesis suggests that patient-provider communication is among the most pressing issues to be targeted for improving healthcare for autistic individuals. This concurs with previous research (6, 7, 16), Thus, developing QI initiatives to improve patient-HCP communication should receive significant attention going forward as this will likely benefit HCPs and patients.

Multi-disciplinary teams

In addition to the studies being inclusive of multiple stakeholder perspectives, this research also benefited from the involvement of a multi-disciplinary team in its development and conduct. Thus, the perspectives of professionals from various disciplines, including psychologists, researchers and physicians working in both primary and secondary care, influenced the research. Multi-disciplinary teams are now recommended for conducting research related to large-scale societal issues, including health services research, which cannot be easily addressed by one discipline (240). The collaboration between researchers and professionals from different disciplines in this instance helped generate interest in this programme of research in various healthcare sectors, including paediatrics and psychiatry, and was particularly helpful for engaging and retaining participants (241). Further, three highly esteemed researchers (Drs Olive Healy and Jennifer Holloway, who are experts in the field of autism-related research, and Prof. Andrew Murphy who is a general practitioner and expert in health services research) were members of my Graduate Research Committee. Their role was to review and guide my work throughout the PhD. The support and expertise offered by the Graduate Research Committee throughout the PhD has ensured the academic and scientific quality of this thesis in line with international standards and facilitated the timely completion of the programme (242, 243).

Outputs

Another strength relates to the tangible outputs resulting from this programme of research. Two novel measurement tools for assessing barriers to care have been developed based on a systematic review of international literature which, after further development and validation, may be adopted into practice, or within organisations, for assessing barriers to care in various settings within the healthcare system. Developing and implementing objective, validated assessments is important for a number of reasons. First, valid and reliable measures are essential for extracting valid and reliable data which can be used to inform QI initiatives (12, 244). Second, standardised, objective measurement tools would allow for bench-marking within services over time and across services in terms of measuring ‘autism-friendliness’ (245). Third, standardised validated measures would be useful for conducting standardised pre and post-test assessments of the effectiveness of interventions or QI initiatives aimed at reducing barriers. Such assessments could allow for meta-analyses to be conducted of particular types of interventions or within particular healthcare settings (246). I was unable to conduct a meta-analysis as part of the systematic review described in Chapter 6 (56) of this thesis because the methods of intervention evaluation across studies were too varied. Thus, the availability of standardised outcome measures which could assess the presence of barriers before and after an intervention would allow for meta-analysis to be conducted.

Foundation on a systematic review

This thesis was founded upon a systematic review of international research on the barriers to care for autistic individuals. Systematic reviews are instrumental in identifying research gaps and informing future evidence-based research; indeed, some researchers posit that no new research should be conducted without first conducting a systematic review if an up-to-date review of the topic does not exist (247-249). Chapter 2 (9) describes the first systematic review to assess barriers to care based on the perspectives of autistic individuals, HCPs, and caregivers; this review provided valuable information and offered a crucial basis for the remainder of the thesis. As discussed earlier, there now exists an abundance of research on the barriers to healthcare that impact on autistic individuals, but there is still a dearth of interventions aimed at reducing barriers and improving care and no systematic review of such interventions had previously been conducted. Thus, a further strength was the inclusion of a second systematic review which has bridged the gap between understanding the barriers experienced by the autism community and the impact these can have, to identifying potential solutions. Although an intervention has not been developed as part of this thesis, Chapter 6 (56) has outlined clear considerations for the next steps to be taken towards developing solutions.

Theoretical considerations

A final strength is that the research was guided by existing conceptualisations and theories of access to healthcare, considering both supply- and demand-side factors of the healthcare system (e.g., 93, 94). This has helped to make the data more actionable by identifying constructs (e.g., accommodation, availability, accessibility) that are important to the autism community and thus should be targeted by intervention (93, 94, 250). The identification of relevant theoretical constructs provides a useful framework for developing any such interventions (251, 252). There are numerous reasons why the application of theory to intervention development is important, including shortening the time needed to develop an intervention, optimising the design, identifying the conditions of context, and promoting transfer of learning from one project to another (253-256). In addition, drawing on exiting theories helps to identify what is important, relevant, and feasible to inform the intended goals, content, and delivery of the intervention (253). Existing theories may also inform the programme theory (i.e., the pathway between the intervention and the expected outcomes) through the use of logic models (253, 257, 258). Thus, the use of theory both strengthens the quality of interventions and facilitates their evaluation (253). Thus, it is hoped that the data produced by this thesis, which had been guided by various access theories, will be useful in guiding future intervention development.

Limitations

Despite the considerable strengths of the work completed, the programme of research also had a number of limitations which must be taken into consideration when interpreting the results. The individual limitations of each of the five studies have been discussed in Chapters 2-6. This section will discuss the limitations of this body of work as a whole. Consideration of these limitations has implications for the interpretation of the findings of this thesis and may inform the planning and conduct of further work in this research area.

Lack of autistic participants

A major limitation of the research programme is the relative lack of the autistic participants in the included studies. The direct involvement of autistic participants in research is important for a number of reasons. First, autistic self-advocates often report that their experiences are minimised or misrepresented by non-autistic parents, researchers, educators, and service providers; thus, the presence of the autistic voice helps to ensure that research topics and experiences are positive and meaningful for autistic individuals (259). Second, autistic individuals can offer the most insight into their own care and thus possess a wealth of information that would advance knowledge and practice (125, 127). Despite substantial efforts to recruit autistic adults to the interview study (Chapter 5), uptake was very low (i.e., just eight autistic adults were recruited). Further work is therefore needed to identify how best to recruit autistic participants to research studies in this area. Such efforts should also consider how to recruit diverse populations of autistic adults, including those or who are considered minimally- or non-speaking or those who have co-occurring IDs (127, 259). Consideration is also needed on how we, as researchers, can best facilitate research participation for these specific sub-populations, who are particularly neglected in self-report research (259).

Lack of participatory involvement in research

A second, related, limitation is the lack of involvement of the autism community in the development of the studies and throughout the research process. Participatory research, defined as incorporating the views of autistic individuals and their supporters about what research gets done, how it is done and how it is implemented (127, 260) is now recommended when conducting autism-related research (125, 128). The benefits of participatory groups in terms of outcomes have been repeatedly demonstrated and if conducted to a high standard, may lead to better translation of research into practice and improved health outcomes for autistic people and their supporters (12, 125, 127). Further, participatory research may facilitate the shift away from a medical model of disability and disablist attitudes, towards a social model of disability (128). This shift is important for empowering autistic individuals to reach their full potential by removing societal imposed challenges (121). Although efforts were made to ensure the current research was aligned with the research priorities outlined by the autism community (1), it was not possible to engage in participatory research due to resource constraints. The landscape in Ireland for participatory research, has changed dramatically in the four years since this research project was first proposed, however. This advancement is demonstrated by the establishment of funded programmes such as the Public and Patient Involvement (PPI) Ignite Network which aims to support researchers and the public to engage in participatory research. The PPI Ignite Network does this by running seminars, workshops, study days and support clinics for researchers, and sharing information about participatory research with the public and organisations (261, 262). It is, therefore, recommended that that going forward, researchers make strong efforts to include the autism community in the entire research process using whatever resources and supports become available to enable and facilitate effective partnerships.

Over-representation of autistic children

A further limitation is that although this research did not aim to assess barriers to care for autistic children only, autistic children are over-represented throughout the studies. In the systematic review of barriers to care (Chapter 2; 9), the majority of studies (*n*=23; 74%) focused on barriers for autistic children. In the caregiver-report tool, caregivers of just 28 (14%) adults participated, all others were parents of children or adolescents. In the interview study, 27 (59%) stories related to autistic children and 19 (41%) related to autistic adults. Finally in the systematic review of interventions, children were participants in 15 (48%) interventions while adults participated in just 3 (10%) interventions. The over-representation of children in studies two (Chapter 3; 10) and four (Chapter 5), was despite efforts to avoid relying solely on recruiting participants through engagement with schools. The over-representation of autistic children in research is common (263, 264), although there has been a definite shift in recent years towards expanding research relating to autistic adults (e.g., 12, 103). Older autistic adults remain very under-represented in research, however (1, 265, 266). The limited research that has investigated health related issues for older autistic adults has demonstrated that many mental and physical health conditions are more prevalent in older autistic adults with and without intellectual disabilities, compared to older adults from the general population (265, 266). In particular, there is a striking lack of research relating to the health of older autistic women which is problematic considering the research which indicates higher rates of health conditions and premature mortality for autistic women (267-269). Many female-specific health risks are associated with reproductive transition points such as puberty and menopause (270-272). Although a small amount of research exists regarding puberty and menstruation for autistic girls and women (273-276), there is almost no research on the effects of the menopause on autistic women (277-279). This is a serious omission as the menopause is known to substantially impact on physical and mental health (280-282). It is, therefore, essential to develop an understanding of the experiences of healthcare pertinent to the older autistic population, giving careful consideration to gender-related health issues, and to develop tailored interventions for improving care delivery. Future research must investigate how to recruit autistic adults of all ages, and such efforts should include caregivers of autistic adults to ensure full representation.

Lack of focus on autistic cohorts

Relatedly, not focusing on one specific cohort of autistic individuals (e.g., adults vs children, individuals with or without co-occurring ID) may also be considered a limitation of this body of work. Focusing on one cohort would have allowed for more direct comparisons to be drawn with other literature focused on just one cohort (e.g., 3, 5) to compare barriers experienced by the different groups. It is to be expected that different sub-populations of the autism community will experience different barriers due to the heterogeneity of the presentation of autism and co-occurring conditions

(103, 283). This is supported by the findings in Chapter 3 (10) that autism severity and co-occurring anxiety were associated with more frequent and severe barriers to care. Thus, future research may want to evaluate the relevance of the taxonomy (Chapter 2; 9) with certain sub-populations of the autism community and assess whether adaptations are required.

Lack of focus on a specific healthcare setting

Similarly, is it possible that this programme of research was too broad in its focus on healthcare as a whole. It may have been more beneficial to focus on one specific area, such as primary care or emergency care, as it is very likely that different barriers will arise in these various settings. For example, the emergency department (ED) may present more sensory challenges than a primary care office due to the larger, busier and more chaotic nature of an ED (26). Indeed, in the systematic review of interventions described in Chapter 6 (56), there were several interventions that were specific to one discipline. For example, Whippey et al. (116) describe the use of a surgical checklist to improve the perioperative experience for autistic children, and Cox et al. (284) describe an intervention aimed at improving tolerance of having an MRI scan. In addition, it is likely that the level of autism knowledge will vary by profession. It may be expected that primary care providers would have more knowledge of autism compared to surgeons for example due to the increased likelihood of contact between primary care physicians and autistic patients (27). Much research has assessed primary care providers’ levels of autism knowledge but there is far less work that has assessed other HCPs levels of autism knowledge (27). This is something future research should consider investigating as this would allow comparisons to be made between professions. Future research may need to refine the tools and taxonomy produced in this thesis to suit the needs of specific disciplines and settings. Nevertheless, consideration of the healthcare system in its entirety may also be considered a strength as the broad overview is a first step to considering the barriers that exist across the system.

Potential for limited generalisability

Finally, with the exception of the two systematic reviews (Chapters 2 and 6) the research is focused on, and reflects, the healthcare experiences of autistic persons in Ireland. Thus, the generalisability of the findings must be considered with caution. The findings from Chapters 3, 4 and 5 were, however, largely congruent with research on barriers to healthcare for autistic individuals conducted in other countries including the UK (e.g., 8, 28), Canada, (e.g., (24, 66), and the USA (e.g., 12, 67). Nevertheless, further validation work on both of the barriers tools presented herein will need to be conducted to assess their usability with international populations and health systems as it has been shown that even relatively robust factor structures can vary in different countries (285). Ireland has a two-tier health system (i.e., a publicly funded health system which operates very closely alongside a privately insured health system), thus, research is also recommended to assess whether the tools could be used in their current forms in other types of health systems, or whether adaptations are required.

Conclusion

People on the autism spectrum experience poorer health outcomes and higher premature mortality rates than the general population. These poor health outcomes are not a necessary consequence of being on the autism spectrum however, rather, they reflect substantial inequities in healthcare whereby autistic individuals have more difficulties accessing the care they need than others. There is, therefore, a pressing need to understand the inequities in, and accessibility of, health for autistic individuals and to begin to consider how these issues can be addressed. Accordingly, the overarching aims of this thesis were to examine the specific barriers to healthcare that autistic individuals experience and to assess the interventions that have been implemented to improve healthcare experiences and overcome barriers for the autistic community. These aims have been addressed comprehensively through completion of a series of five studies, utilising both qualitative and quantitative methods. These studies have identified key barriers that exist at the level of the patient (e.g., communication/social issues), HCP (e.g., a lack of autism knowledge) and system (e.g., a lack of coordination between services/HCPs). Further, the findings from Chapter 6 (56) indicate that interventions that have been designed and trialled to date show little evidence of user involvement in design and tend to focus on the patient (i.e., help the autistic person to tolerate, cooperate with a medical procedure). There has been less focus on changing the behaviour of HCPs or making changes at the level of the system/organisation, despite the prevalence of HCP and system-related barriers which have been indicated by autistic individuals, caregivers, and HCPs.

In addressing these aims, this thesis produced a number of deliverables. In Chapter 2 (9), a taxonomy of barriers to healthcare was developed which may allow for the development of future assessment tools and guide intervention development. Chapter 3 (10) described the development and evaluation of a novel caregiver-report Barriers to Healthcare Tool, while Chapter 4 described the development and evaluation of a novel physician report Barriers to Providing Healthcare Tool. Chapter 5 provided insight into how barriers can impact on the care experiences and health of autistic individuals. An additional strength of the thesis was the overview of existing interventions which was provided in Chapter 6 (56). This thesis, therefore, constitutes an important foundation for future work on health inequity and healthcare for autistic individuals. It is clear that future research must begin mapping barriers to interventions and ensure to involve autistic individuals in all stages of intervention development. In practice, healthcare professionals need to begin to use the available tools to systematically assess the barriers to care that exist within their settings and to consistently implement reasonable adjustments to ensure that autistic individuals have adequate access to high quality care. Finally, Ireland needs to follow in the footsteps of other EU countries and the UK and develop a National Autism Strategy for improving access to, and delivery of, services to the autism community and this must include a specific focus on improving physical health and healthcare. Current prevalence estimates attest that one in every 160 people are estimated to be autistic, and this number is rising (286). Thus, it is imperative that our models of healthcare delivery change in order to meaningfully support inclusion and ensure optimal health for the entire autistic community.

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Appendices

Appendix One

1.1 Search Strategy Medline.

	Search term
1	Exp autistic disorder/
2	Exp autism spectrum disorder/
3	Exp asperger syndrome/
4	Exp developmental disabilities/
5	Exp intellectual disabilities/
6	Exp child development disorders, pervaisive/
7	Autis*.ti,ab
8	ASD.ti,ab
9	Asperger*.ti,ab
10	“Development* dis*”.ti,ab
11	“learning dis*”.ti,ab
12	“intellectual dis*”.ti,ab
13	1 OR 2 OR 3 OR 4 OR 5 OR 6 OR 7 OR 8 OR 9 OR 10 OR 11 OR 12
14	Exp primary health care/
15	Exp family practice/
16	Exp general practitioners/
17	Exp physicians/
18	Exp medical staff, hospital/
19	Exp secondary care/
20	Exp hospitals/
21	Exp general practice/
22	Exp Nurses/ or nurse administrator/ or exp nurse practitioner/ or exp nurse specialists/ or exp nurses, community health/ or exp nurses, international/ or exp nurses, male/ or exp nurses, public health/ or exp nursing staff/ or exp nursing staff, hospital/
23	Healthcare.ti,ab
24	GP.ti,ab
25	Physician*.ti,ab
26	Doctor*.ti,ab
27	“General pract*”.ti,ab
28	“Medical staff”.ti,ab
29	Hospital*.ti,ab
30	Nurs*.ti,ab

31	“primary *care”.ti,ab
32	“Family pract*”.ti,ab
33	“Secondary *care”.ti,ab
34	“emergency *care”.ti,ab
35	“Emergency room*”.ti.ab
36	“Emergency department*”.ti,ab
37	14 or 15 OR 16 OR 17 OR 18 OR 19 OR 20 OR 21 OR 22 OR 23 OR 24 OR 25 OR 26 OR 27 OR 28 OR 29 OR 30 OR 31 OR 32 OR 33 OR 34 OR 35 OR 36
38	Barrier*.ti.ab.
39	Fit.ti.ab.
40	“reasonable adjustment*”.ti.ab
41	*equity.ti.ab
42	Exp healthcare disparities/
43	Exp health services accessibility/
	38 OR 39 OR 40 OR 41 OR 42 OR 43
44	13 AND 37 AND 43
45	Limit 44 to English
46	Limit 45 to peer-reviewed

EXP=explode; ti=title, ab=abstract;

1.2. Characteristics of Included Studies and Quality Appraisal Scores.

First author, Location	Study design	Participants (n)	Sampling method	Healthcare setting	Barriers reported/endorsed	QATSDD score
Ahmedani & Hock (45); USA	Cross-sectional survey	Caregivers nos* (*n* not specified) of children (*n*=1,427)	Sample taken from larger study (NSCH) which used random sampling	Not specified	• Out of pocket expenses are not reasonable; • Benefits do not cover insurance; • Being less satisfied with between-provider communication; • Providers not listening to parents; • Providers not being sensitive to family values; • Not feeling like a partner in child's care;	21
Benich et al. (49); USA	Qualitative interviews	Mothers (*n*=10); Father (*n*=1); Grandmother (*n*=1) of autistic children	Convenience sampling	Secondary care: surgery; otorhinolaryngology	• Waiting times; • Loud noises & sudden movements; • Communication issues; • Too many interactions with healthcare staff; • Changing from daily routine;	17
Bultas (62); USA	Qualitative interviews	Mothers (*n*=11) of preschool-age autistic children	Convenience sampling	Primary care	• HCPs unable to use knowledge of autism to make accommodations; • HCPs do not understand that delivery of healthcare must be adjusted for autistic children; • Not listening to the expertise of the mother; • Not acknowledging caregiver's need to look at alternative treatments; • If there was no medication to prescribe to solve or cure the problem, the primary care provider was not interested in addressing the problem; • Child behaviours; • Wait times;	24

					• Waiting room set-up; • Lack of distraction/toys available in waiting room; • HCP office did not provide adequate environment for the physical resources necessary to meet child's needs;	
Bultas et al. (60); USA	Cross-sectional survey	Paediatric physicians (*n*=27); Paediatric nurses (*n*=25); barriers based on 49 responses); Parents nos (*n*=58); barriers based on 45 responses)	Convenience sampling	Office based settings	HCPs: • Child behaviour during encounter; • Uncooperative, unable to completely examine; • Child fears about the examination; • Difficulty communicating with child; • Parent overwhelmed during encounter; • Child fears about the provider; • Parent distracted by child; • Family dynamics; • Parent beliefs about healthcare; • Office environment; Parents: • HCP/staff doesn't understand impact of autism; • Child behaviour during encounter; • Environmental issues (e.g., waiting, lights, noises); • Fast pace of office; • Child communication deficits; • Need case management for child; • Parent distracted during encounter; • Severe disconnect between different aspects and professionals involved in child's care;	17

Carbone et al. (81); USA	Pre/Post intervention surveys	Physicians (*n*=43)	Convenience sampling	Primary care	• Lack of care coordination resources; • Lack of practice guidelines; • Lack of time during visits; • Lack of provider education about autism; • Patient's frequent use of complimentary alternative medicine; • Patient's frequent use of outside providers; • Families being hesitant about or refusing vaccines; • Lack of community resources;	19
Chiri & Warfield (46); USA	Cross-sectional survey	Parents nos (*n* not specified) of autistic children (*n*=2,088)	Screened sample from larger study which used random sampling	Routine preventative care; specialty care; therapy services; mental healthcare;	Health plan-based access problems • Costs too much; • No insurance; • Health plan problem; • No referral; • Provider does not accept insurance; Provider based access problems • Not available in area/transportation; • Inconvenient times; • Provider did not know how to treat; • Dissatisfaction with provider; • Did not know where to go.	19
Davignon et al. (59); USA	Qualitative interviews	Mothers (*n*=20); Nurses (*n*=8); Physicians (*n*=4); Nurse Practitioner (*n*=3); Child life specialist	Convenience sampling	Tertiary care	• Training often inadequate to understand how to approach and communicate with autistic children; • Lack of systems to support knowledge of individual child's needs and challenges; • Inconsistent information sharing between staff; • Preparatory guidance received by parents is variable;	24

		(*n*=3); Medical assistant (*n*=2)			• Feeling rushed; • Long wait times in waiting room; • Not recognising need for modifications;	
Dern et al. (44); Germany	Qualitative: two collaborative meetings & review of practical guidelines on managing autistic patients– no other information provided	Autistic adults and professionals working in field of autism – no other information provided (*n* not specified)	Convenience sampling	Not specified	Making appointments • Difficulties making phone calls/emails/fax/letters; Waiting area • Physical closeness to other patients while waiting; • Stress due to uncertainty of length of wait; • Sensory overstimulation through visual stimuli; • Disturbing sounds can be perceived as discomforting, painful/irritating, can have exhaustive effects (e.g., sirens); Examination • Discomfort due to unannounced touch; • Reduced pain perception or untypical behaviour when having painful experience; • Context and consequences, beginning and ending of an examination unclear; Communication • Stress due to open questions; • Lack of time to think and respond; • Lack of appreciation of written notes; • General difficulties in verbal and nonverbal communication; • Mimic, gesture and prosody possibly limited or do not match metal state or meaning (e.g., pain);	6

					• Difficulties identifying, generalising, naming, localising and reporting sensory impressions and observations of inner experience (e.g., pain); • Peculiar language and images (idiosyncratic of hyper specific); • Literal understanding of language; • Lack of initiative to report medical and related issues; • Not providing essential information if not directly asked; Hospital • Stress due to staff change; • Not admitting to the hospital because of rigid thinking e.g., watering plants at home and not seeing an alternative for that; Sensory issues • Prosopagnosia (facial recognition); • Anxiety due to not recognising medical staff; • Difficulties and fear of sensory overload when travelling via public transport or stress due to sirens or overall sound volume;	
Eseigbe et al. (67); Nigeria	Cross-sectional survey	Medical doctors (*n*=167); Barriers reported by 19 participants	Convenience sampling	Primary care; secondary care; tertiary care.	• Dearth of speech and behavioural therapists; • High cost of patient evaluation; • Poor understanding and acceptance of autism by caregivers; • High default rate from follow-up care;	19
Golnik et al. (71); USA	Cross sectional survey	Primary care physicians (*n*=539):	Simple random sampling	Primary care	• Lack of care coordination; • Lack of reimbursement; • Family is sceptical of traditional medicine;	23

		(Paediatricians (*n*=449); Family physicians (*n*=90))			• Family is sceptical of vaccines; • Patient's frequent use of complementary alternative medicine; • Patient's use of outside providers; • Lack of practice guidelines; • Lack of time during office visit; • Lack of provider education about autism;	
Krauss et al. (47); USA	Cross sectional survey	Caregivers (*n* not specified) of autistic children (*n*=152)	Sample taken from larger study -data not available	Specialty medical care	Health plan-based access problems • Getting referrals; • The health plan would not pay; • Getting the number of visits needed; • The amount the family had to pay; Provider based access problems • Getting appointments; • Finding skilled and experienced specialty doctors; • Coordination of care between specialty doctor and other providers;	18
Kuhlthau et al. (69); USA	Cross-sectional survey	Mothers (*n*=164) & other caregivers nos(*n*=19) of autistic youth	Convenience sampling	Primary care	• Lack of information on the transition process (child to adult care); • Difficulty finding an adult primary care provider who is sufficiently knowledgeable about autism; • Difficulty finding an adult primary care provider who is autism-friendly or willing to accept child as patient; • Difficulty finding an adult medical specialist who is autism-friendly or willing to accept child as patient;	20

					• Lack of coordination and communication between paediatrician and adult primary care provider; • Difficulties related to insurance coverage; • Difficulties related to guardianship;	
Lake et al. (52); Canada	Qualitative focus groups	Mothers (*n*=7) of autistic adolescents or adults	Convenience sample taken from larger study which used purposive sampling	Not specified	• Lack of physician expertise; • Not satisfied with support/assistance from physician; • Prescribing healthcare professionals did not refer them to specialist and could not provide non-pharmacological services; • Prescribing healthcare professionals did not take enough time to really understand and get to know their child and therefore could not properly address or monitor their needs; • Poor communication between doctors, patients, and parents – physician's failure to acknowledge parents' opinions concerns and suggestions. • Healthcare professionals reluctant to take on primary responsibility for monitoring medication effects and for ensuring that medications were used in the most efficient way. • Difficulty finding and accessing essential services and supports; • Services and supports time limited when they were accessed; • Little integration within healthcare system (e.g., transitions from paediatric to adult care was difficult:	23

					• Communication lacking with healthcare professionals resulting in poor consistency of services, unnecessary redundancy, and overall lower quality of service;	
Lindberg et al. (64); Sweden	Qualitative interviews	Mothers (*n*=7) & Fathers (*n*=5) of autistic children	Convenience sampling	Surgery/ anaesthetics	• Having to repeatedly relate their concerns; • New personnel who lack understanding of why child behaves in such a way; • Lack of continuity during the perioperative process; • Too many unfamiliar people unendurable for the child; • Lack of support from HCP; • Contact between child and staff during perioperative process were too sporadic; • Unfinished dialogues between parents and staff due to interruptions (e.g., phone calls/staff change-overs); • Interventions interrupted by phone calls/staff changeovers; • Lack of time to help the child; • Lack of knowledge to help the child; • Misguided routines that govern perioperative care; • Intrusive glances from other parents in the waiting room;	18
Lum et al. (70); Australia	Cross-sectional survey	Autistic adult women (*n*=32); non-autistic women (*n*=26)	Convenience sampling	General and maternity healthcare	General healthcare • Waiting rooms caused anxiety; • Anxiety caused in healthcare settings caused autistic participants to experience greater	16

					reductions in their capacity to communicate verbally; Maternity healthcare • Information and support during pregnancy were problematic for autistic women; • Communication of pain and needs during childbirth were more problematic for autistic women; Autism stigma and disclosure • 'I felt disclosure would affect my treatment or communication';	
Mazurek et al. (68); USA	Pre/post intervention surveys	Primary care paediatricians (*n*=10); Family medical physician (*n*=1); Nurse (*n*=2); other (*n*=1)).	Purposive sampling	Primary care	• Lack of time; • Lack of access to autism specialists; • Lack of self-efficacy in managing autistic children; • Lack of support from administration; • Inadequate reimbursement;	20
Muskat et al. (55); Canada	Qualitative interviews	Autistic Youth (*n*=6); Mothers (*n*=19) & Fathers (*n*=3); Nurses (*n*=6); physicians (*n*=6); SLP	Convenience sampling	Secondary care	Communication • Challenges with respect to youth understanding language and expressing themselves; Sensory processing • Inundated with sensory experiences in the hospital (e.g., touching). Challenges working with the healthcare team; • Many HCPs in the room at the same time can be overwhelming for child;	28

		(*n*=1); Social worker (*n*=1)			Waiting/transitioning • Waiting and transitioning through the hospital/changing rooms during the stay; • Staff inflexibility; • Rigidity of hospital;	
Nicholas et al. (61); Canada	Focus groups (*n*=60)	Parents nos (*n*=16); HCPs (*n*=44): (administrators (*n*=6); Family centered care council member (*n*=1); Healthcare trainee (*n*=2; Nurse (*n*=4); OT (*n*=3); physician (*n*=15); psychometrist (*n*=3); researcher (*n*=6); social worker (*n*=2) SLP (*n*=1); undisclosed (*n*=1).	Sample taken from earlier study which used convenience sampling	Emergency Department	• Unstructured waiting in the emergency department; • Too many interactions with healthcare staff; • HCP fear of not knowing how to manage aggressive behaviour; • Substantial resource gaps in autism support and related health service systems impede appropriate emergency department follow-up; • Parents may not be as welcomed or engaged with in non-paediatric facilities because of a heightened orientation to individual patient privacy and a presumption of adult agency;	28
Nicholas et al. (53); Canada	Qualitative interviews	Mothers (*n*=24) & Fathers (*n*=7);	Convenience sample	Emergency Department	• Children overwhelmed when having to move between areas of hospital; • Sensory issues (e.g., lights, being touched);	21

		Autistic children with (*n*=4)			• Wait times for beds – remaining in sensory-heightened emergency department environment; • Gaps in follow-up care; • Children unable to report source of their discomfort; • Children having difficulty comprehending that painful procedures are for their benefit; • Parents feel criticised by HCPs due to bad behaviour of child; • Lack of knowledge of effective ways to support autistic children; • Staff communicated in ways that were ineffective for the child and caused more anxiety; • Doctors in the emergency department ignored the child despite the child asking many questions; • Medical jargon incomprehensible to child; • Staff unprepared to effectively address autism-related challenges; • Parents not sufficiently consulted about their child’s emergency department care; • Parents feeling dismissed by staff in emergency department; • Lengthy wait times in emergency department; • Restraints used on child so procedures could be completed as quickly as possible; • Needles cause anxiety;	

Nicolaidis et al. (57); USA	Qualitative interviews	Autistic adults (*n*=39); Caregivers nos (*n*=16).	Maximum variation sampling	Not specified	Autism-related patient level factors • Verbal communication skills (not knowing how to respond to questions from staff); • Sensory sensitives; • Challenges with body awareness (not knowing how to describe pain); • Slow processing speed; • Need for consistency; • A-typical non-verbal communication; • Challenges with organisation; Provider level factors • HCP lack of knowledge about autism in adults (can over attribute behaviours to autism); • Providers incorrect assumptions about individual patients' skills/needs; • Providers' unwillingness to allow patients to communicate in writing; • Failure to use accessible language; • lack of skill in appropriately incorporating supporters; System level factors • Lack of formal/informal supports; • Accessibility of healthcare settings (loud sensory stimulating waiting rooms); • Stigma about autism; • Complexity of healthcare system;	27
Okumura et al. (72); USA	Cross sectional survey about 1 of 2	Paediatricians (*n*=1163)	Convenience sampling	Primary and non-primary paediatrics	Primary care (autism vignette) • Lack of administrative time to make phone calls/coordinate care/follow on referrals/review patients' charts etc.;	24

	vignettes (1 on child with chronic medical condition, 1 on an autistic child).				• Lack of time for office visit; • Lack of administrative staff support to make phone calls/coordinate care/follow on referrals etc.; Non-primary care (autism vignette) • Lack of administrative time; • Lack of staff support;	
Raymaker et al. (41); USA	Cross sectional survey	Autistic adults (*n*=209); Adults with disabilities (*n*=55); Adults no autism, no disabilities (*n*=173)	National convenience sample	Not specified	Emotional • Fear/anxiety; • Embarrassment; • Worry that stress of interacting with healthcare system will cause patient to lose control: • Frustration/anger; • Lack of confidence; • Fatigue/pain; Executive function • Trouble following up on care; • Often miss appointments due to memory problems; • Trouble following medical instructions the way they are presented; • Difficulty understanding how to translate medical information into concrete steps that can be taken to improve health; Healthcare navigation • Don’t understand the healthcare system/find it too hard to work through. – e.g., managed care, billing system;	26

					• Too hard to seek primary care/follow-up with primary care; • Problems filling out paperwork; Provider attitudes • Patient behaviours misinterpreted; • Staff don't believe patient when told that new symptoms are not related to existing condition/disability; • Staff do not take patient's communication seriously; • Staff unwilling to communicate in the mode patient has specified; • Can't find provider who will accommodate patient's needs; Patient-provider communication • Unable to process information fast enough to participate in real-time discussions about healthcare; • Difficulty communicating with doctors/staff; • Trouble following spoken directions; • Appointments too short to accommodate communication needs; • Difficulty moving/communicating effectively when in crisis; • Difficulty identifying and reporting pain/other physical symptoms; Sensory • Healthcare facilities cause sensory discomfort;	

					• Sensory issues make it difficult to communicate in healthcare settings; • Sensory issues make tests, screenings, and medical exams difficult/impossible; Socio-economic • Concern about cost of care; • Don’t have insurance coverage; • Insurance doesn’t cover care coordination services; • Trouble getting reimbursements for atypical treatments; • Transportation costs too much; • Living in rural area/doctor’s office too far away; • Co-payments are too high; • Waiting for insurance plan approval; Waiting • Hard to handle the waiting room; • Wait in healthcare office is too long;	
Russell & McClonsky (50); USA	Mixed methods: Qualitative interviews and survey	Mothers (*n*=11) of autistic children	Purposive and snowball sampling	Primary care	• Waiting rooms – unfamiliar people and surroundings, judgement from other parents/support staff if child misbehaved; • Parents’ expertise not respected to by HCP; • Parent concerns ignored; • Parents views not sought; • Child not treated with dignity; • Not feeling they could approach the HCP for information/advice about child’s disability;	26
Saqr et al. (48);	Mixed methods:	Survey: (*n*=112) not	Convenience sampling	Primary care	Survey	22

USA	Pre-visit telephone assessment survey; qualitative focus groups;	broken down by group: caregivers nos & autistic persons; Focus groups: autistic adults (*n*=10)			• Difficulty in the waiting room/waiting in general; • Not liking noises; • Aversion to needles; • Difficulty being touched; • History of aggression in medical facility; • Not liking bright lights; • Being unable to tolerate vital signs; Focus groups • Difficulties communicating with the physician; • The physical exam; • The waiting room experience; • Anticipation of social interaction with the medical staff created stress; • Anxiety and overstimulation inhibit ability to focus and communicate well with medical staff;	
Strunk et al. (54); USA	Qualitative interviews	Mothers (*n*=10) & Fathers (*n*=2) of autistic adolescents	Voluntary and snowball sampling	Not specified	• Inadequate healthcare services; • No advocacy for their adolescents’ healthcare needs; • Physicians not interested in what parents had to say; • Physicians too busy to listen to parents; • A lack of medical personnel specifically qualified to work with autistic adolescents; • Often felt rushed during their adolescents’ appointments; • No consistency/collaboration between physical and mental healthcare services;	24

					• Financial burden; • Parents feeling they were perceived negatively by doctors; • Adolescents negatively treated; • Challenges managing specific medical procedures;	
Unigwe et al. (51); UK	Cross sectional survey with 1 qualitative open-ended question.	GPs (*n*=304)	Convenience, purposive and snowball sampling	Primary care	• Lack of clear referral pathways; • Long waiting lists; • Limited resources; • Lack of joined up services; • Support for autistic adults virtually non-existent; • Lack of support for families; • Limited support from local services; • Lack of clarity over GP remit; • Consultations too short; • Need for specific training on autism;	26
Van Cleave et al. (63); USA	Qualitative interviews	Paediatricians (*n*=7); Specialists (*n*=12); Nurses (*n*=4); Front desk (*n*=1); Mothers (*n*=5) & Fathers (*n*=1) of autistic children;	Purposive & snowball sampling	Primary care and specialty care	HCPs: • Interpreting symptoms complicated by autism-related behaviours; • Difficult to develop strategies to carry out diagnostic procedures and treatment recommendations that account for autism-related behaviours; • No reliable source for specialty communication; • Coordination among primary care providers and specialists was challenging;	26

					• Can be difficult to figure out which specialist to contact; • Need for more staff training, resources, strategies about how to approach autistic children from a behavioural standpoint; Parents • Communication between PCPs and Specialists seems uncommon; • Waiting in the waiting room; • Inconsistencies across visits;	
Warfield et al. (66); USA	Qualitative interviews	Physicians (*n*=9); Nurse (*n*=1)	Convenience & purposive sampling	Primary care	System level • Shortage of medical and non-medical services and support for youth and autistic adults; • A general lack of providers willing to work with autistic adults; • Financial disincentives due to work-related issues of time, reimbursement, and the need for additional staff; • Fewer services available for adults than for autistic children; • Lack of needed coordination and infrastructure on the adult medical side; • Challenges getting services for autistic youth if they do not also have a diagnosis of intellectual disability; Practice/provision level • Time constraints; • Organisational issues; • Complexity of family involvement;	21

					• Physical environments not appropriately designed; • Communicating with patients during visit; • Autistic persons can have different treatment needs which present challenges; • The individualised nature of the condition of autism makes standardising care difficult; • Lack of knowledge about autism care, services in the community and others who can help with service provision; • General complexity of care needs of autistic individuals; Training and education level • Lack of formal education/training in medical school/residency; • Lack of knowledge around working with autistic individuals;	
Will et al. (65); USA	Cross sectional survey	Nurses (*n*=126)	Convenience sampling	Primary care	• Lack of care coordination; • Family is sceptical of vaccines; • Lack of time during offices visit; • Lack of provider education about autism; • Lack of practice guidelines; • Lack of reimbursement; • Patients’ frequent use of complimentary alternative medicine; • Patients’ use of outside providers; • Family is sceptical of traditional medicine;	22
Zerbo et al. (58); USA	Mixed methods: Cross	Survey: Physicians	Survey: convenience sampling;	Primary care and secondary care	• Poor knowledge and training; • Difficulty communicating with patients with limited verbal abilities;	26

	sectional survey; Qualitative interviews	(*n*=593); Nurses (*n*=58); Interviews: Physicians (*n*=9)	Interview: purposive sampling		• Need for improvement in transfer of care from paediatrics to adult medicine; • Office visits longer for autistic patients when other family members present; • Issues regarding conservatorship and privacy when carers accompany patient;	
Zwaigenbaum et al. (56); USA	Qualitative interviews	Physicians (*n*=10); Nurses (*n*=12)	Convenience sampling	Emergency Department	Characteristics of the child • Aggressiveness; • Extreme symptom severity; • Limited verbal communications; • Children have difficulty coping with lengthy waiting times; Emergency department environment • Sensory triggers; • Rigid procedures to follow in hospital – move quick and efficiently – not suitable for autistic children; • Unfamiliarity of environment and HCPs; • Too many HCPs can be overwhelming for child; Competing demands in emergency department • Resource constraints/need to balance other patients' needs;	20

GP=general practitioner; HCP=healthcare provider; nos=not otherwise specified; PCP=primary care provider

Appendix Two

2.1. Comparison of Observed Eigenvalues with Eigen Values Generated from the Parallel Analysis

Eigenvalue #	Extracted Eigenvalue	% of variance	Cumulative %	Random percentile Eigenvalue
1	13.099	31.88	31.88	2.14
2	3.361	8	39.19	1.99
3	2.269	5.40	44.6	1.88
4	1.840	3.5	62.48	1.80
5	1.471	3.59	52.36	1.74
6	1.416	3.37	55.85	1.65
7	1.204	2.87	58.72	1.59
8	1.132	2.7	61.41	1.53
9	1.024	2.44	63.85	1.48
10	1.018	2.43	66.28	1.43
…				
42	.122	.29	100	0.35

2.2. Process of item deletion through the EFA

2.2.1. First Iteration of EFA

Survey items	**Factor 1**	**Factor 2**	**Factor 3**	**Factor 4**
Child/adult finds appointments overwhelming;	.837			
Child/adult finds it hard to handle the wating area;	.722			
Child/adult dislikes doctors;	.718		-.219	
Child/adult finds it difficult to tolerate medical procedures;	.681			
Healthcare facilities cause child/adult sensory discomfort;	.659			
Caregiver is afraid to take child/adult to doctor;	.608			.277
Caregiver finds appointments overwhelming;	.605			.340
Caregiver worries that stress of interacting with healthcare system will cause child/adult distress;	.595	-.258	.304	
Child/adult has difficulty identifying/reporting pain/symptoms	.558			
Child/adult has difficulty communicating their thoughts/wants/needs;	.525			
The wait in healthcare settings is too long;	.399		.249	
Healthcare providers are unwilling to communicate in patient's preferred communication modality;	.391		.229	
Pain prevents the autistic person from going to the doctor;	.370			.277
There are language barriers;	.353	.289		
Insurance coverage impacts access to care;		.747		
Cost of care impacts access to care;		.696		
Transport costs too much;		.687		.208
Insurance does not cover needed care;		.675		
There are other transportation problems;		.644		
Caregiver does not know where to go;		.600		
There is inadequate caregiver/family support;		.551		
There is no doctor available when needed;		.495	.280	
Social isolation impacts access to care;		.487		.312
The healthcare facility is not open at suitable times;		.477		
Caregivers disagree over healthcare;	.268	.470		
Consultations are too short;		.436	.308	
Getting appointments can be difficult;		.434	.304	
There are physical accessibility problems;	.275	.372		
The waiting list for services is a problem;		.228		
Healthcare providers do not listen;			.824	
The child/adult's behaviours are misinterpreted by healthcare providers;			.750	
Caregiver does not like how healthcare providers have treated the child/adult in the past;			.745	

Healthcare providers have inadequate knowledge/education for treating autistic people;			.686	
Caregiver knows others who have had negative healthcare encounters;	-.201		.617	
Finding a healthcare provider who will accommodate the child/adult needs can be difficult;	.202		.606	
Caregiver finds it difficult to remember to attend appointments;				.598
Medical recommendations given to the child/adult can be hard to follow;				.583
Following up on child/adult’s care can be difficult				.485
The treatment plan can be hard to follow;	.291			.463
Finding time to go to the doctor can be difficult;		.363		.379
Going to medical appointments can be hard to fit in;		.341		.359
Labelling/stigma can be a problem;				.228

2.2.2. Second Iteration of EFA

Survey items	**Factor 1**	**Factor 2**	**Factor 3**	**Factor 4**
Insurance coverage impacts access to care;	.764			
Cost of care impacts access to care;	.432			
Transport costs too much;	.693			.244
Insurance does not cover needed care;	.686			
There are other transportation problems;	.650			
Caregiver does not know where to go;	.590			
There is inadequate caregiver/family support;	.547			.222
There is no doctor available when needed;	.493		.289	
Social isolation impacts access to care;	.488		-.206	.365
Caregivers disagree over healthcare;	.457	.226		
The healthcare facility is not open at suitable times;	.445			
Consultations are too short;	.440		.304	
Getting appointments can be difficult;	.432		.295	-.217
Child/adult finds appointments overwhelming;		.854		
Child/adult finds it difficult to tolerate medical procedures;		.717	-.223	
Child/adult finds it hard to handle the wating area;		.710		
Child/adult dislikes doctors;		.684	-.220	
Healthcare facilities cause child/adult sensory discomfort;		.634		
Caregiver worries that stress of interacting with healthcare system will cause child/adult distress;	-.288	.610	.303	
Caregiver finds appointments overwhelming;		.583		.304
Child/adult has difficulties identifying/reporting pain/symptoms;		.581		
Caregiver is afraid to take child/adult to doctor;		.576		.252
Child/adult has difficulty communicating their thoughts/wants/needs;		.534		
Healthcare providers do not listen;			.809	
The child/adult’s behaviours are misinterpreted by healthcare providers;			.754	
Caregiver does not like how healthcare providers have treated the child/adult in the past;			.714	
Healthcare providers have inadequate knowledge/education for treating autistic people;			.676	
Finding a healthcare provider who will accommodate the child/adult’s needs can be difficult;			.612	
Caregiver knows others who have had negative healthcare encounters;		-.236	.610	
Medical recommendations given to the child/adult can be hard to follow;				.680
The treatment plan given to the child/adults can be hard to follow;		.213		.560

Following up on child/adult’s care can be difficult;	.516
Caregiver finds it difficult to remember to attend appointments;	.511

2.2.3. Third Iteration of EFA

Survey items	**Factor 1**	**Factor 2**	**Factor 3**	**Factor 4**
Child/adult finds appointments overwhelming;	.853			
Child/adult finds it difficult to tolerate medical procedures;	.726		-.220	
Child/adult finds it hard to handle the wating area;	.703			
Child/adult dislikes doctors;	.688		-.238	
Healthcare facilities cause child/adult sensory discomfort;	.631		.204	
Caregiver worries that stress of interacting with healthcare system will cause child/adult distress;	.601	-.212	.308	
Caregiver finds appointments overwhelming;	.587			.283
Caregiver is afraid to take child/adult to doctor;	.583			.249
Child/adult has difficulties identifying/reporting pain/symptoms;	.577			
Child/adult has difficulty communicating their thoughts/wants/needs;	.532			
Insurance coverage impacts access to care;		.805		
Cost of care impacts access to care;		.747		
Insurance does not cover needed care;		.728		
Transport costs too much;		.718		.237
There are other transportation problems;		.637		
Caregiver does not know where to go;		.541		
There is inadequate caregiver/family support;		.525		
Caregivers disagree over healthcare;	.236	.449		
There is no doctor available when needed;		.440	.264	
The healthcare facility is not open at suitable times;		.400		
Healthcare providers do not listen;			.816	
The child/adult's behaviours are misinterpreted by healthcare providers;			.800	
Healthcare providers have inadequate knowledge/education for treating autistic people;			.721	
Caregiver does not like how caregivers have treated the child/adult in the past;			.699	
Finding a healthcare provider who will accommodate the child/adult's needs can be difficult;			.631	
Caregiver knows others who have had negative healthcare encounters;	-.234		.614	
Medical recommendations given to the child/adult can be hard to follow;				.698
The treatment plan can be hard to follow;	.216			.587
Caregiver finds it difficult to remember to attend appointments;				.552
Following up on child/adult's care can be difficult;				.524

2.2.4. Fourth Iteration of EFA

Survey items	**Factor 1**	**Factor 2**	**Factor 3**	**Factor 4**
Child/adult finds appointments overwhelming;	.854			
Child/adult finds it difficult to tolerate medical procedures;	.723		-.219	
Child/adult finds it hard to handle the wating area;	.707			
Child/adult dislikes doctors;	.683		-.237	
Healthcare facilities cause child/adult sensory discomfort;	.632		.201	
Caregiver worries that stress of interacting with healthcare system will cause child/adult distress;	.605	-.215	.301	
Caregiver is afraid to take child/adult to doctor;	.588			.254
Caregiver finds appointments overwhelming;	.587			.285
Child/adult has difficulties identifying/reporting pain/symptoms;	.579			
Child/adult has difficulty communicating their thoughts/wants/needs;	.530			
Insurance coverage impacts access to care;		.808		
Cost of care impacts access to care;		.746		
Transport costs too much;		.724		.231
Insurance does not cover needed care;		.721		
There are other transportation problems;		.626		
Caregiver does not know where to go;		.530		
There is inadequate caregiver/family support;		.529		
Caregivers disagree over healthcare;	.235	.439		
The healthcare facility does not open at suitable times;		.388		
Healthcare providers do not listen;			.827	
The child/adult’s behaviours are misinterpreted by healthcare providers;			.809	
Healthcare providers have inadequate knowledge/education for treating autistic people;			.724	
Caregiver does not like how healthcare providers have treated the child/adults in the past;			.706	
Finding a healthcare provider who will accommodate the child/adult’s needs can be difficult;			.636	
Caregiver knows others who have had negative healthcare encounters;	-.236		.620	
Medical recommendations given to the child/adult can be hard to follow;				.699
The treatment plan can be hard to follow ;	.216			.587
Caregiver finds it difficult to remember to attend appointments;				.555

Following up on child/adult’s care can be difficult;	.527

2.2.5. Fifth Iteration of EFA

Survey items	**Factor 1**	**Factor 2**	**Factor 3**	**Factor 4**
Child/adult finds appointments overwhelming;	.849			
Child/adult finds it difficult to tolerate medical procedures;	.727		-.220	
Child/adult finds it hard to handle the wating area;	.700			
Child/adult dislikes doctors;	.680		-.232	
Healthcare facilities cause child/adult sensory discomfort;	.628		.206	
Caregiver worries that stress of interacting with healthcare system will cause child/adult distress;	.602	-.205	.298	
Caregiver is afraid to take child/adult to doctor;	.591			.248
Caregiver finds appointments overwhelming;	.583			.288
Child/adult has difficulties identifying/reporting pain/symptoms;	.577			
Child/adult has difficulty communicating their thoughts/wants/needs;	.533			
Insurance coverage impacts access to care;		.834		
Cost of care impacts access to care;		.754		
Insurance does not cover needed care;		.731		
Transport costs too much;		.703		.247
There are other transportation problems;		.579		
There is inadequate caregiver/family support;		.528		
Caregiver does not know where to go;		.479		
Caregivers disagree over healthcare;	.233	.402		
Healthcare providers do not listen;			.826	
The child/adult's behaviours are misinterpreted by healthcare providers;			.812	
Healthcare providers have inadequate knowledge/education for treating autistic people;			.725	
Caregiver does not like how healthcare providers have treated the child/adult in the past;			.701	
Finding a healthcare provider who will accommodate the child/adult's needs can be difficult;			.635	
Caregiver knows others who have had negative healthcare encounters;	-.228		.615	
Medical recommendations given to the child/adult can be hard to follow;				699
The treatment plan can be hard to follow ;	.216			.583
Caregiver finds it difficult to remember to attend appointments;				.557
Following up on child/adult's care can be difficult				.532

2.2.6. Final Iteration of EFA

Survey items	**Factor 1**	**Factor 2**	**Factor 3**	**Factor 4**
Child/adult finds appointments overwhelming;	**.841**			
Child/adult finds it difficult to tolerate medical procedures;	**.725**		-.218	
Child/adult finds it hard to handle the wating area;	**.697**			
Child/adult dislikes doctors;	**.674**		-.225	
Healthcare facilities cause child/adult sensory discomfort;	**.624**		.206	
Caregiver worries that stress of interacting with healthcare system will cause child/adult distress;	**.601**		.292	
Caregiver is afraid to take child/adult to doctor	**.586**			.250
Caregiver finds appointments overwhelming;	**.584**			.287
Child/adult has difficulties identifying/reporting pain/symptoms;	**.582**			
Child/adult has difficulty communicating their thoughts/wants/needs;	**.537**			
Insurance coverage impacts access to care;		**.825**		
Cost of care impacts access to care;		**.759**		
Insurance does not cover needed care;		**.727**		
Transport costs too much;		**.692**		.254
There are other transportation problems;		**.562**		
There is inadequate caregiver/family support;		**.513**		
Caregiver does not know where to go;		**.454**		
Healthcare providers do not listen;			**.825**	
The child/adult’s behaviours are misinterpreted by healthcare providers;			**.810**	
Healthcare providers have inadequate knowledge/education for treating autistic people;			**.722**	
Caregiver does not like how healthcare providers have treated the child/adult in the past;			**.705**	
Finding a healthcare provider who will accommodate the child/adult’s needs can be difficult;			**.635**	
Caregiver knows others who have had negative healthcare encounters;	-.222		**.614**	
Medical recommendations given to the child/adult can be hard to follow;				**.697**
The treatment plan can be hard to follow;	.214			**.582**
Caregiver finds it difficult to remember to attend appointments;				**.556**
Following up on child/adult’s care can be difficult				**.530**

2.3. Appendix Final Barriers to Healthcare Tool

Please indicate the barriers/difficulties you have experienced by circling the number in the appropriate boxes. Please provide <u>**two**</u> answers to <u>**each**</u> statement.

- **(A)** Please choose from the <u>left-hand</u> column (1-4) to indicate **how often** that barrier typically occurs **(frequency)**.

- **(B)** Please choose from the <u>right-hand</u> column (1-3) to indicate **how much of a problem** that barrier represents (**severity**).

If the barrier is not an issue, circle 'never' (i.e. number 0).

Please indicate the barriers you have experienced in the **last 12 months**.		**(A) Frequency**				**(B) Severity**		
	Never	**Rarely**	**Sometimes**	**Often**	**Very Often**	**Slight**	**Moderate**	**Severe**
Patient-level barriers								
1. S/he finds it difficult to tolerate medical procedures.	0	1	2	3	4	1	2	3
2. S/he finds appointments overwhelming.	0	1	2	3	4	1	2	3
3. S/he dislikes doctors.	0	1	2	3	4	1	2	3
4. I (caregiver) find appointments overwhelming.	0	1	2	3	4	1	2	3
5. I'm (caregiver) afraid to take him/her to the doctor.	0	1	2	3	4	1	2	3
6. I (caregiver) worry that the stress of interacting with the healthcare system will cause him/her to become distressed.	0	1	2	3	4	1	2	3
7. S/he has difficulties identifying and reporting pain and or/other symptoms.	0	1	2	3	4	1	2	3
8. Healthcare facilities cause him/her sensory discomfort (e.g., the lights, smells or sounds make visits uncomfortable).	0	1	2	3	4	1	2	3
9. S/he finds it hard to handle the waiting room.	0	1	2	3	4	1	2	3
10. S/he has difficulty communicating their thoughts, wants or needs.	0	1	2	3	4	1	2	3
HCP-level barriers	0	1	2	3	4	1	2	3
1. The healthcare providers do not listen.	0	1	2	3	4	1	2	3
2. His/her behaviours are misinterpreted by healthcare providers.	0	1	2	3	4	1	2	3
3. Healthcare providers have inadequate knowledge/education on treating persons with ASD.	0	1	2	3	4	1	2	3

4. I (caregiver) do not like how healthcare providers have treated him/her in the past.	0	1	2	3	4	1	2	3
5. Finding a healthcare provider who will accommodate his/her needs (e.g., dim lighting/quiet room) can be difficult.	0	1	2	3	4	1	2	3
6. I (caregiver) know others who have had negative experiences with healthcare services.	0	1	2	3	4	1	2	3
System-level barriers								
1. Insurance impacts our access to healthcare.	0	1	2	3	4	1	2	3
2. Cost of care impacts our access to healthcare.	0	1	2	3	4	1	2	3
3. Insurance does not cover the care we need.	0	1	2	3	4	1	2	3
4. Transport costs too much.	0	1	2	3	4	1	2	3
5. There are other transportation problems.	0	1	2	3	4	1	2	3
6. Inadequate social, family or caregivers support is a problem.	0	1	2	3	4	1	2	3
7. I (caregiver) don’t know where to go.	0	1	2	3	4	1	2	3
Barriers related to managing care								
1. It can be hard to remember to attend appointments.	0	1	2	3	4	1	2	3
2. Following up on his/her care can be difficult.	0	1	2	3	4	1	2	3
3. Medical recommendations given to him/her can be hard to follow.	0	1	2	3	4	1	2	3
4. The treatment plan (e.g., give medicine/take temperature) given to him/her can be hard to follow.	0	1	2	3	4	1	2	3

ASD=autism spectrum disorder; HCP=healthcare provider

2.4 Pearson correlation tables

2.4.1 Pearson's Correlations Between Predictor Variables and the Total Frequency of Barriers

	Total frequency of barriers	Autistic person's age	Autistic person's gender	Autistic person's health status	GP visits	Routine hospital visits	ED visits	Autism severity	Depression	Anxiety	OCD	GAP	Unmet needs
Total frequency of barriers;	1												
Autistic person's age;	-.234**	1											
Autistic person's gender;	-.069	-.010	1										
Autistic person's health status;	-.210**	-.089	.014	1									
GP visits;	.229**	-.052	-.082	-.261***	1								
Routine hospital visits;	.157*	-.120	-.181**	-.167*	.177**	1							
ED visits;	.221**	-.104	.072	-.143*	-.314***	.141*	1						
Autism severity;	.498***	.009	.029	-.148*	.128*	.102*	.086	1					
Depression;	.438***	.048	-.089	-.384***	.226**	.045	.174**	.445***	1				
Anxiety;	.444***	.072	-.018	-.314***	.223**	.081	.166*	.511***	.708***	1			
OCD;	.327***	.014	.089	-.182**	.141*	-.003	.095	.426***	.476***	.557***	1		
GAP;	.608***	-.051	-.043	-.222**	.262***	.065	.013*	.641***	.642***	.655***	.538***	1	
Unmet needs;	.295***	-.005	-.093	-.152*	.216**	.138*	.159*	.109	.215**	.234**	.174**	.244***	1

**p*<0.05; ***p*<0.01; ****p*<0.001; GP=General Practitioner; ED=Emergency Department; OCD=Obsessive Compulsive Disorder; GAP=General Adjustment Problems

2.4.2. Pearson's Correlations Between Predictor Variables and the Frequency of Patient-Level Barriers

	Total frequency of patient-level barriers	Autistic person's age	Autistic person's gender	Autistic person's health status	GP visits	Routine hospital visits	ED visits	Autism severity	Depression	Anxiety	OCD	GAP	Unmet needs
Total frequency of patient-level barriers;	1												
Autistic person's age;	-.016*	1											
Autistic person's gender;	-.01	-.051	1										
Autistic person's health status;	-.045	-.089	.104	1									
GP visits;	.109	-.052	-.082	-.261***	1								
Routine hospital visits;	.075	-.12	-.181**	-.167*	.177**	1							
ED visits;	.182**	-.104	.072	-.143*	.341***	.141*	1						
Autism severity;	.551***	-.009	.029	-.148*	.128*	.102	.086	1					
Depression;	.332***	-.048	-.089	-.384***	.226**	.045	.174**	.445***	1				
Anxiety;	.341***	-.072	-.108	-.314***	.223**	.081	.166*	.511***	.708***	1			
OCD;	.25***	-.114	.089	-.182**	.141*	-.003	.095	.426***	.467***	.557***	1		
GAP;	.577***	-.051	-.043	-.222**	.262***	.065	.13*	.641***	.642***	.655***	.583***	1	
Unmet needs;	.092	-.005	-.093	-.152**	.216**	.138*	.159*	.109**	.215**	.234**	.174**	.244***	1

**p*<0.05; ***p*<0.01; ****p*<0.001; GP=General Practitioner; ED=Emergency Department; OCD=Obsessive Compulsive Disorder; GAP=General Adjustment Problems

2.4.3. Pearson's Correlations Between Predictor Variables and the Frequency of Healthcare Provider-Level Barriers

	Total frequency of HCP-level barriers	Autistic person's age	Autistic person's gender	Autistic person's health status	GP visits	Routine hospital visits	ED visits	Autism severity	Depress ion	Anxiety	OCD	GAP	Unmet needs
Total frequency of HCP-level barriers;	1												
Autistic person's age;	.016*	1											
Autistic person's gender;	-.105	-.01	1										
Autistic person's health status;	-.196**	-.089	.104	1									
GP visits;	.165*	-.052	-.082	-.261***	1								
Routine hospital visits;	.144*	-.012	-.181**	-.167*	.177**	1							
ED visits;	.139*	-.104	.072	-.143*	.341***	.141*	1						
Autism severity;	.331***	.009	.029	-.148*	.128*	.102	.086	1					
Depression;	.31***	.048	-.089	-.384***	.226**	.045	.174**	.445***	1				
Anxiety;	.287***	.072	-.108	-.314***	.223**	.081	.166*	.511***	.708***	1			
OCD;	.184**	.014	.089	-.182**	.141*	-.003	.095	.426***	.467***	.557***	1		
GAP;	.428***	-.051	-.043	-.222**	.262***	.065	.13*	.641***	.642***	.655***	.583***	1	
Unmet needs;	.279***	-.005	-.093	-.152*	.216**	.138*	.159*	.109**	.215**	.234**	.174**	.244***	1

*$p<0.05$; **$p<0.01$; ***$p<0.001$; HCP=Healthcare provider; GP=General Practitioner; ED=Emergency Department; OCD=Obsessive Compulsive Disorder; GAP=General Adjustment Problems

2.4.4. Pearson's Correlations Between Predictor Variables and the Frequency of Healthcare System-Level Barriers

	Total frequency of System-level barriers	Autistic person's age	Autistic person's gender	Autistic person's health status	GP visits	Routine hospital visits	ED visits	Autism severity	Depression	Anxiety	OCD	GAP	Unmet needs
Total frequency of system-level barriers;	1												
Autistic person's age;	-.202**	1											
Autistic person's gender;	-.057	-.01	1										
Autistic person's health status;	-.239**	-.089	.104	1									
GP visits;	.251***	-.052	-.082	-.261***	1								
Routine hospital visits;	.150*	-.012	-.181**	-.167*	.177**	1							
ED visits;	.179**	-.104	.072	-.143*	.341***	.141*	1						
Autism severity;	.356***	.009	.029	-.148*	.128*	.102	.086	1					
Depression;	.382***	.048	-.089	-.384***	.226**	.045	.174**	.445***	1				
Anxiety;	.451***	.072	-.108	-.314***	.223**	.081	.166*	.511***	.708***	1			
OCD;	.318***	.014	.089	-.182**	.141*	-.003	.095	.426***	.467***	.557***	1		
GAP;	.475***	-.051	-.043	-.222**	.262***	.065	.13*	.641***	.642***	.655***	.583***	1	
Unmet needs;	.323***	-.005	-.093	-.152*	.216**	.138*	.159*	.109**	.215**	.234**	.174**	.244***	1

*$p<0.05$; **$p<0.01$; ***$p<0.001$; GP=General Practitioner; ED=Emergency Department; OCD=Obsessive Compulsive Disorder; GAP=General Adjustment Problems

2.4.5. Pearson's Correlations Between Predictor Variables and the Frequency of Barriers Related to Managing Care

	Frequency of barriers related to managing care	Autistic person's age	Autistic person's gender	Autistic person's health status	GP visits	Routine hospital visits	ED visits	Autism severity	Depression	Anxiety	OCD	GAP	Unmet needs
Frequency of barriers related to managing care	1												
Autistic person's age;	-.199**	1											
Autistic person's gender;	-.062	-.01	1										
Autistic person's health status;	-.205**	-.089	.104	1									
GP visits;	.184**	-.052	-.082	-.261***	1								
Routine hospital visits;	.136*	-.012	-.181**	-.167*	.177**	1							
ED visits;	.156*	-.104	.072	-.143*	.341***	.141*	1						
Autism severity;	.287***	.009	.029	-.148*	.128*	.102	.086	1					
Depression;	.308***	.048	-.089	-.384***	.226**	.045	.174**	.445***	1				
Anxiety;	.220**	.072	-.108	-.314***	.223**	.081	.166*	.511***	.708***	1			
OCD;	.236**	.014	.089	-.182**	.141*	-.003	.095	.426***	.467***	.557***	1		
GAP;	.378***	-.051	-.043	-.222**	.262***	.065	.13*	.641***	.642***	.655***	.583***	1	
Unmet needs;	.162*	-.005	-.093	-.152*	.216**	.138*	.159*	.109**	.215**	.234**	.174**	.244***	1

*p<0.05; **p<0.01; ***p<0.001; *GP=General Practitioner; ED=Emergency Department; OCD=Obsessive Compulsive Disorder; GAP=General Adjustment Problems*

2.4.6. Pearson's Correlations Between Predictor Variables and the Total Severity of Barriers

	Total severity of barriers	Autistic person's age	Autistic person's gender	Autistic person's health status	GP visits	Routine hospital visits	ED visits	Autism severity	Depression	Anxiety	OCD	GAP	Unmet needs
Total severity of barriers	1												
Autistic person's age;	-.221**	1											
Autistic person's gender;	-.009	2015	1										
Autistic person's health status;	-.264**	-.062	-.001	1									
GP visits;	.250**	-.046	-.078	-.324***	1								
Routine hospital visits;	.161*	-.112	-.154*	-.153*	.134	1							
ED visits;	.209*	-.047	-.094	-.255**	.334***	.235**	1						
Autism severity;	.498***	.005	-.09	-.234**	.015	.082	.223**	1					
Depression;	.449***	.078	-.061	-.368***	.271**	-.002	.266**	.458***	1				
Anxiety;	.446***	.098	-.067	-.332***	.17*	-.001	.186*	.469***	.701***	1			
OCD;	.316***	.126	.166*	-.244**	.05	-.015	.089	.382***	.489***	.568***	1		
GAP;	.642***	-.057	.041	-.252**	.204*	.03	.219**	.659***	..649***	.641***	.558***	1	
Unmet needs;	.210*	.035	-.068	-.102	.128	.098	.125	.147	.157*	.189*	.108	.013	1

*$p<0.05$; **$p<0.01$; ***$p<0.001$; GP=General Practitioner; ED=Emergency Department; OCD=Obsessive Compulsive Disorder; GAP=General Adjustment Problems

Appendix Three

Appendix 3.1 Two factor solution with extracted factors, items, internal consistency, and variance explained

Item	Factor 1	Factor 2
Factor 1: Patient-related factors; Cronbach's α=0.9; variance explained 38.9%		
The patient's reactivity to the healthcare environment	**.830**	-.169
There are insufficient resources	**.752**	.038
Consultations are too short to accommodate patients on the autism spectrum	**.744**	-.034
There are communication difficulties	**.724**	-.044
Lengthy waiting room times for patients on the autism spectrum	**.701**	-.048
Challenging behaviours exhibited by the patient	**.655**	-.034
Limited flexibility to accommodate patients on the autism spectrum and their needs	**.629**	.183
There is a lack of coordination between services	**.569**	.157
The patients' use of outside providers (e.g., alternative therapies)	**.506**	.190
The physical environment is unsuitable for patients on the autism spectrum	**.503**	.190
Factor 2: Provider and system-related factors; Cronbach's α=0.8; Variance explained: 6.0%		
There is a lack of clarity regarding GP remit/referral	-.011	**.742**
There is a lack of providers willing to work with patients on the autism spectrum	.008	**.733**
There is a lack of guidelines for working with autistic patients	-.028	**.716**
I prefer not to work with patients on the autism spectrum	-.170	**.627**
The patient's family is sceptical of conventional medicine (e.g., vaccines)	.074	**.516**
Family/carer involvement makes provision of healthcare for patients on the autism spectrum more complex	.101	**.513**
There are financial disincentives due to the need for additional time with the patient.	.196	**.505**
Lack of own knowledge regarding autism.	.053	**.478**

GP=general practitioner;

Appendix 3.2 Additional items that were removed during analysis

	Item
1	There is a lack of resources
2	There is a lack of access to autism specialists
3	There is limited flexibility to accommodate patients on the autism spectrum
4	There is a lack of guidelines for caring for people on the autism spectrum
5	Lack of confidence in own ability to care for people on the autism spectrum

Appendix 3.3. Summary of regression analyses

Variable	***F change***	R^2	*A* R^2
Variables associated with frequency of patient-related barriers			
1. No. of autistic patients seen per annum	1.633	.008	.003
2. Autism knowledge Medical specialty Years since graduation Previous autism training	.883	.043	-.002
Variables associated with frequency of HCP/family-related barriers			
1. No. of autistic patients seen per annum	.953	.005	.000
2. Autism knowledge Medical specialty Years since graduation Previous autism training	1.735	.072	.028
Variables associated with frequency of system-related barriers			
1. No. of autistic patients seen per annum	.214	.001	-.004
2. Autism knowledge Medical specialty Years since graduation Previous autism training	1.626	.064	.021

HCP=healthcare provider

Appendix 3.4. Final Barriers to Providing Healthcare measurement tool

		Frequency					Severity		
For each item, circle the number in the appropriate boxes to indicate: **(A) how often** the described barrier typically occurs for you when providing healthcare to a patient on the autism spectrum; (*frequency*); **AND** **(B) how much of a problem** that barrier represent for you (*severity*) If the barrier is not a problem for you, circle 'never' (i.e., '0') and move to the next item.		Never	Rarely	Sometimes	Often	Very often	Slight	Moderate	Severe
1	Challenging behaviours exhibited by the patient.	0	1	2	3	4	1	2	3
2	There are communication difficulties.	0	1	2	3	4	1	2	3
3	The patient's reactivity to the healthcare environment.	0	1	2	3	4	1	2	3
4	Lengthy waiting room time for patients on the autism spectrum.	0	1	2	3	4	1	2	3
5	The patients' use of outside providers (specialists, chiropractors, therapists etc.).	0	1	2	3	4	1	2	3
6	Consultations are too short to accommodate patients on the autism spectrum.	0	1	2	3	4	1	2	3
7	There is a lack of clarity regarding GP remit/referral.	0	1	2	3	4	1	2	3
8	There are financial disincentives due to the need for additional time with the patient.	0	1	2	3	4	1	2	3
9	The patient's family is sceptical of conventional medicine (e.g., vaccines).	0	1	2	3	4	1	2	3
10	These is a lack of providers willing to work with patients on the autism spectrum.	0	1	2	3	4	1	2	3
11	Family/carer involvement makes healthcare provision for patients on the autism spectrum more complex.	0	1	2	3	4	1	2	3
12	I prefer to avoid working with patients on the autism spectrum.	0	1	2	3	4	1	2	3

13	There is a lack of support for patients and caregivers.	0	1	2	3	4	1	2	3
14	The physical environment in healthcare settings is unsuitable for patients on the autism spectrum.	0	1	2	3	4	1	2	3
15	Lack of own knowledge regarding autism	0	1	2	3	4	1	2	3
16	There is a lack of coordination between services.	0	1	2	3	4	1	2	3
17	There are shortages of medical and non-medical services for people on the autism spectrum.	0	1	2	3	4	1	2	3

GP=general practitioner; *Note*: Due to large amounts of missing data, no analyses were conducted using the severity scale; The EFA is based on frequency scale data only.

Appendix Four

Appendix 4.1 Online Supplemental Digital Content 1- Medline Search Strategy

1. Autism*.ti,ab.
2. Asperger*.ti,ab.
3. “development* dis*”.ti,ab.
4. Exp autistic disorder/
5. Exp autism spectrum disorder/
6. Exp Asperger syndrome/
7. Exp developmental disabilities/
8. 1 OR 2 OR 3 OR 4 OR 5 OR 6 OR 7
9. Exp primary health care/
10. Exp family practice/
11. Exp physicians/
12. Exp medical staff/
13. Exp secondary care/
14. Exp hospitals/
15. Exp general practice/
16. GP.ti,ab.
17. “Primary *care”.ti,ab.
18. “Family pract*”.ti,ab.
19. “general pract*”.ti,ab.
20. “secondary *care”.ti,ab.
21. Hospital*.ti,ab.
22. “emergency *care”.ti,ab.
23. “emergency department*”.ti,ab.
24. “emergency room*”.ti,ab.
25. Physician*.ti,ab.
26. Doctor*.ti,ab.
27. “medical care*”.ti,ab.
28. Nurse*.ti,ab.
29. 9 OR 10 OR 11 OR 12 OR 13 OR 14 OR 15 OR 16 OR 17 OR 18 OR 19 OR 20 OR 21 OR 22 OR 23 OR 24 OR 25 OR 26 OR 27 OR 28
30. Interven*.ti,ab.
31. Train*.ti,ab.
32. Toolkit*.ti,ab.
33. Teach*.ti,ab.
34. Program*.ti,ab.
35. Module*.ti,ab.
36. 30 OR 31 OR 32 OR 33 OR 34 OR 35
37. 8 AND 29 AND 36
38. Limit 37 to English

Appendix 4.2 Online Supplemental Digital Content 2 - Study-by-Study Summary of Included Studies

Study	Country	Setting	Participants	Research approach	Nature of the Intervention	Measurement	Outcomes	QATSDD Score*
Birkan et al. (55)	USA and Turkey	**Educational Setting for Autistic Children/Adults** *Detail:* School classroom	**Autistic Children/Adults** *Detail:* 6 Autistic children (5M, 1F) ranging in age from 5 years to 16 years	**Quantitative-SSRD** *Detail:* Multiple changing criterion designs	**Patient-focused** *Target:* Cooperation with injections ----- **Behavioural intervention drawn from Science of ABA** *Description:* Behavioural intervention (drawn from science of ABA)-skills teaching, token economy and reinforcement	**Level 3- Behaviour** *Detail:* Measured correct responses on the task in absence of stereotypy and disruptive behaviour.	**Level 3-Behaviour: Positive** *Detail*: Error rates were low during intervention; After completing the program, all 6 participants received injections with only 1 showing distress after injection; Behaviour was observed to generalise to other instructors and settings; Behaviour maintained during post-intervention probes	**14**
Broder-Fingert et al. (56)	USA	**Secondary Care** *Detail:* Hospital	**Autistic Children/Adults; Caregivers of Autistic People** *Detail:* 92 families with an autistic child	**Quantitative-Group research** *Detail:* Non-random comparison study	**Provider-focused** *Target:* To support hospital staff to make individual accommodations in the provision of healthcare for autistic people. -----	**Level 1- Reactions** *Detail:* Assessment of experience with the plan, ease of use, perceived utility, experience of care, beliefs about staff treatment of parents and child likelihood of	**Level 1-Reactions: Positive** *Detail*: The plan was believed to be well tolerated by parents and	**21**

					Care Plan *Description:* An autism specific care plan for autistic children developed through a quality improvement program with questions to address: expressive and receptive communication, social and pragmatic concerns, and safety. It was available online for patient to complete, and then uploaded onto the patient's electronic record.	recommending the hospital, rating of overall experience, staff's attention to autism-related needs and autism-specific concerns during the visit. **Level 4A: Measures of Results (Change in Organisational Practice)** *Detail:* Length of stay and whether there was a safety report or not for 33 patients with a plan as compared to 109 without a plan.	used frequently by staff. Parents with a plan reported better hospital experience and staff attention to their autistic child's specific needs than those without a plan. **Level 4A- Measures of Results (Change in Organisational Practice): No clear effect** *Detail:* There was no difference in length of stay or number of safety reports	
Burnham Riosa et al. (78)	Canada	**Secondary Care** *Details:* Paediatric Hospital	**Healthcare Providers** *Detail:* 102 healthcare professionals and other hospital staff (although 325 staff completed module).	**Quantitative- Group research** *Detail:* Post-test only	**Provider-focused** *Target:* Increase knowledge of hospital staff about autism. ------ **Educational intervention for Staff** *Description:* online training module for healthcare	**Level 1: Reactions** *Detail*: Survey on ease of completion, knowledge gained, helpfulness of info and recommend to colleague	**Level 1- Reactions: Positive** *Detail:* 88% perceived information from module helpful in daily work, and 100% agreed or strongly agreed	**15**

					professionals. Training module took 10 minutes and covered introduction to autism and recommended proactive and reactive strategies for caring for paediatric autistic patients. After completing the training, the participants then applied the knowledge gained to two cases studies. 80% score in quiz to pass the training.		module easy to complete, and 63% agreed/strongly agreed they learned something new.	
Carbone et al. (57)	USA	**Primary Care** *Detail:* Primary care practices	**Healthcare Providers; Caregivers of Autistic People; Healthcare Administrators** *Detail:* 20 paediatric and 6 family medicine teams. Each practice team consisted of at least 1 physician, a nurse or medical assistant, an office manager, and a parent of an autistic child.	**Quantitative-Group research** *Detail:* Pretest/ Posttest	**Provider-focused** *Target:* Physician's self-efficacy in providing care to autistic children. ----- **Quality Improvement**: Detail: The intervention included a training workshop (topics such as care for an autistic child, the challenges faced in meeting healthcare needs, and QI and the use of PDSA cycles). Then, teams developed plans for change (e.g., increasing awareness	**Level 2A: Learning (Changes in Attitudes)** *Detail:* Physician self-efficacy- level of confidence in addressing Autism-specific needs and Autism-associated conditions along with perceived barriers to providing care for autistic children. **Level 4B: Measures of Results (Changes in the Organisation)** *Detail:* Subjective ratings of progress towards achieving	**Level 2A-Learning (Changes in Attitude): No clear effect** *Detail:* Physician self-efficacy improved on all 7 Autism-specific needs and all 9 Autism-associated conditions; Ratings of barriers to care provision decreased significantly for	**12**

					of autism among parents and staff; improving chronic care management of autistic children) and timeframe. To support implementation of plans, there were monthly conference calls on Autism-related topics and two site visits from a QI specialist.	family cantered care goals	two of eight barriers **Level 4B- Measures of Results (Changes in the Organisation): No clear effect** *Detail:* Average rating of progress was 6.5 on a scale of 1 to 10.	
Cavalari et al. (58)	USA	**Educational Setting for Autistic Children/Adults** *Detail:* Nursing suite at a day and residential school for children and youths with	**Autistic Children/Adults** *Detail*: One autistic 16-year-old (F) with an intellectual disability.	**Quantitative- SSRD** *Detail:* Changing Criterion Design	**Patient-focused** *Target:* Compliance with medical examination ----- **Behavioural intervention drawn from science of ABA** *Detail:* Prerequisite skills training; Then, graduated exposure	**Level 3- Behaviour** *Detail:* Number of steps in medical examination hierarchy completed correctly	**Level 3- Behaviour: Positive** *Detail:* Clear improvements in behaviour from phase to phase as criterion was changed; Generalisation	**13**

		Intellectual and Developmental Disabilities			and positive reinforcement, which included a social story and in-vivo modelling.		of behaviour to nursing staff.	
Chebuhar et al. (59)	USA	**Secondary Care** *Details:* Tertiary hospital	**Healthcare Providers** **Caregivers of Autistic People** *Details:* Eight members of staff and nine parents/care givers	**Quantitative- Group research** *Detail:* Post-intervention only	**Patient-focused** *Target:* To make expectation of a hospital visit clear to autistic patients ----- **Picture Schedules** *Description:* Use picture schedules to inform autistic patients of what is expected during visit.	**Level 1- Reactions** *Detail:* Survey for staff and parents to rate the child's maladaptive behaviour, comfort, and feasibility of using the picture schedule.	**Level 1- Reactions: Positive** *Detail:* The majority of staff and caregivers (>75%) thought the picture schedule reduced anxious behaviour of children and led to parents exhibiting less distress. 50% of parents/caregivers thought that they would be less anxious about the next appointment. 75% of staff though the picture schedule made it easier to complete the task or procedure, and 100% were willing to use the picture schedule.	**16**

Clark et al. (60)	UK	**Home and Community** *Details:* Community learning disability service	**Healthcare Administrators** *Details:* 13 administrative staff	**Mixed Methods** *Detail:* Post-intervention only	**Provider-focused** *Target:* Knowledge of autism of administrative staff to allow them to make changes in their practice in order to better accommodate autistic patients. ----- **Educational intervention for healthcare staff** *Description:* introduction to autism training session: didactic teaching covering social interactions, communication, sensory processing issues, repetitive behaviour, cognitive aspects, and ways to support autistic people. Case vignettes to support the generation of goals for changing administrative practice with autistic people.	**Level 1: Reactions** *Detail:* Focus group with eight administrators who had attended the training. Addressed things they liked/disliked about the training, things they had learned. **Level 4A: Measures of Results (Changes in Organisational Practices)** *Detail:* Changes they had made to practices as a result of the training.	**Level 1- Reactions: Positive** *Detail:* Participants were very positive about the training and provided tips for future training. **Level 4A- Measures of Results (Changes in Organisational Practices): Positive** *Detail:* Participants spoke about positive changes to administrative practice in the areas of communication and changes to the waiting areas.	**14**
Cox et al. (61) Study 1	Canada	**Unclear** *Detail:* Unspecified Rectangular	**Autistic Children/Adults** *Detail:* Seven autistic children	**Quantitative: SSRD** *Detail:* Non-concurrent	**Patient-focused** *Target:* Approach mock MRI scanner	**Level 3: Behaviour** Detail: Step completion on stimulus fading sequence	**Level 3- Behaviour: Positive**	**24**

		room equipped with a mock scanner	(5M, 2F), 5 of whom had a co-morbid intellectual disability, ranging in age from 5.5 – 14 years.	multiple baseline across participants design.	and lie in the bore for five minutes ----- **Behavioural intervention drawn from science of ABA**. *Detail:* Stimulus fading/graduated reinforcement, and prompting		*Detail:* Three of the participants demonstrated mastery during baseline (stimulus fading alone). The remaining four participants received the intervention and all achieved mastery.	
Cox et al. (61) Study 2	Canada	**Unclear** *Detail:* Unspecified Rectangular room equipped with a mock scanner	**Autistic Children/ Adults** *Detail:* Five autistic children (2F, 3M) ranging in age from 7-14 years	**Quantitative-SSRD** *Detail:* Multiple baseline design across participants.	**Patient-focused** *Target:* Spend longer periods of motion control in the MRI scanner ----- **Behavioural intervention drawn from science of ABA** *Detail:* Progressive differential reinforcement procedure and prompting	**Level 3: Behaviour** *Detail:* Head motion-steadiness	**Level 3-Behaviour: Positive** *Detail:* One participant achieved mastery during baseline conditions. The four remaining participants all achieved mastery during the intervention phase; Two of the three participants performed at mastery level during the follow-up; Three of the five participants in study 2 subsequently	**24**

							completed a real MRI exam successfully.	
Cuvo et al. (62)	USA	**Educational setting for Autistic Children/Adults; Health Profession Education; Home and Community** *Detail:* Autism centre, Medical office, and child's bedroom	**Autistic Children/ Adults** *Detail:* Six autistic children (5M, 1F; 2 diagnosed with PDD-NOS) ranging in age from 3 to 6 years.	**Quantitative- SSRD** *Detail:* Multiple probe across responses design	**Patient-focused** *Target:* Compliance with physical examination ----- **Behavioural intervention drawn from science of ABA** *Detail:* video modelling, contact desensitisation, shaping, prompting, differential reinforcement of other behaviour, and escape extinction.	**Level 3: Behaviour** *Detail:* Compliance with components of the medical exam; Problem Behaviour	**Level 3- Behaviour: Positive** *Detail:* All participants achieved compliance with all targeted medical exams; Behaviour was observed to generalise to a different setting and different staff members; Behaviour was maintained at 1 month follow-up	**28**
Davit et al. (63)	USA	**Other** *Details:* Clinical research setting	**Autistic Children/ Adults** **Caregivers of Autistic People** *Detail:* 58 families were enrolled in the intervention group; 239 in the control group	**Quantitative- Group research** *Detail:* Quasi randomised controlled trial	**Patient-focused** *Target:* Compliance with venepuncture ----- **Behavioural Intervention drawn from Cognitive Behavioural Therapy** *Description:* A blood draw intervention program was developed based on cognitive behavioural therapy. It consisted of	**Level 3: Behaviour** *Detail:* Blood draw compliance. A successful blood draw was defined as a sufficient sample to send for genetic analysis	**Level 3- Behaviour: Positive** *Detail:* Increased blood draw compliance from 85.4% to 96.6%.	**19**

					(1) instructions for parents on how to practice with their child; (2) instructions for the child; (3) individualised social story integrating a parent chosen reward as a motivator; (4) boardmaker picture schedule; and (5) packet of venepuncture materials			
Ellis et al. (64)	USA	**Home and Community** *Detail:* Participant's home	**Autistic Children/Adults** *Detail:* One 4-year-old autistic child (M)	**Quantitative SSRD** *Detail:* Changing criterion design combined with a multiple baseline across stimuli design.	**Patient-focused** *Target:* Tolerance of skin care products including antibiotic cream ----- **Behavioural intervention drawn from science of ABA** *Detail:* graduated exposure, contingent attention, and modelling.	**Level 1- Reactions** *Detail:* Measure of social validity through which parents rated the goal and methods, and the satisfaction with the method and outcome. **Level 3- Behaviour** *Detail:* Accepting responses and rejecting responses measured.	**Level 1- Reactions: Positive** *Detail:* Goal and method rated as very important, and satisfaction rated as very satisfied. Parents can now apply antibiotic cream to scrapes which they couldn't before, and which got infected. No suggestions for improvement and the parents saw very quick results.	**23**

							Level 3- Behaviour: Positive Participant learned to tolerate the application of the lotion; Behaviour persisted at 1 month follow-up and generalised to application of cream by mother.	
Ghuman et al. (65)	USA	**Unclear**	**Autistic Children/Adults** *Detail:* Four autistic children with comorbid symptoms associated with ADHD that were aged between 5 and 6.5 years and with IQ of 50 or above; 1 child withdrew from the study	**Quantitative- Group research** *Detail*-Pilot feasibility study (post-test only)	**Patient-focused** *Target:* Pill swallowing skills taught. ----- **Behavioural intervention drawn from science of ABA** *Detail:* pre-requisite skills training, verbal instruction, in vivo modelling, prompting, graduated exposure/shaping, and reinforcement.	**Level 3- Behaviour** *Detail:* Percentage of the following behaviours during session- Acceptance, Swallow; Chew; Expel; Avoidance.	**Level 3- Behaviour: Positive** *Detail:* One child withdrew after four sessions when goal was not achieved although progress was documented; Prior to training none of the children could successfully swallow the pills. Notable increase in the percentage of pills accepted and swallowed by all 4 children	**9**

							– up to 95%, 96.2%, 100% and 67.4% of pills accepted; One child learned to swallow the study capsules with the behaviour therapist but didn't perform the behaviour with the parent.	
Giarelli et al. (66)	USA	**Educational Setting for Autistic Children/Adults** *Details:* Education facility at regional autism treatment centre	**Healthcare Providers** *Detail:* 37 participants. 92% female, 35% nurses worked in adult care, 30% nurses working as administrators.	**Quantitative-Group research** *Detail:* Post-intervention only	**Provider-focused** *Target:* Knowledge of nurses about autism. ----- **Educational Intervention for healthcare staff** *Description:* Continuing education program for nurses. Two day workshops covering prevalence, aetiology, risk factors, early identification, and comorbid medical psychiatric and behavioural conditions, evidence-based treatment/interventions , effect of Autism on individual and family, educational and healthcare services	**Level 1: Reactions.** *Detail:* Course evaluation survey. Survey on how the participants used the knowledge gained from the training, did they share the knowledge with colleagues.	**Level 1-Reactions: Positive** *Detail:* All participants valued the course content. 74% shared information with employers, and 94% with co-workers. 77% incorporated learning into practice, and 35% intended to educate peers following course.	**14**

					available in the community, and nursing care of autistic individuals.			
Gillis et al. (67)	USA	**Educational Setting for Autistic Children/Adults** *Detail:* Specialised school for autistic children	**Autistic Children/ Adults** *Detail:* 18 Autistic children (15M, 3F), ranging in age from 2.7-13.8 years.	**Quantitative-Group research** Details: Specific research design unclear but data reported at group level and comparison between moderate and very good progress groups conducted.	**Patient-focused** *Target:* Address fear of routine physical exams. ----- **Behavioural intervention drawn from science of ABA** *Detail:* reinforced practice, modelling and graduated exposure using a fear hierarchy.	**Level 3- Behaviour** *Detail:* Behavioural Observation Assessment used which allowed for rating of approach and avoidance behaviours.	**Level 3-Behaviour: Positive** *Details:* 15 of 18 participants met the criterion of having a neutral or positive rating of behaviour during exposure to the entire physical exam across two sessions, 1 week apart; Progress was made but intervention was not completed for 3 participants due to pragmatic reasons associated with research in a school setting; Results appeared to be maintained at follow-up an average of 8 months later although four participants still	**26**

							showed fear at follow-up (3 who had completed and 1 who had not completed).	
Golnik et al (68)	USA	**Primary Care** *Detail:* Primary care medicine in an autism specific medical home	**Autistic Children/Adults** *Detail:* 46 autistic children in the intervention group, 157 autistic controls who received standard medical care. 34 intervention group and 62 control group completed both surveys	**Quantitative-Group research** *Detail:* Quasi experimental pre-test/post-test control design	**Patient-focused** *Target:* Experience of primary care for autistic children ------ **Quality Improvement** *Description:* Autism-specific medical home intervention using a quality improvement approach. Autism-related accomplishments included: Autism care plan, change monitoring log, coordinating with outside resources, tools to improve appointments, ASD specific toys, and longer duration visits.	**Level 2A: Learning (Changes in Attitudes)** *Detail:* Questionnaire for caregivers regarding access, usage, satisfaction, unmet needs, and family stress.	**Level 2A: Learning (Changes in Attitudes): No clear effects** *Detail:* Compared to controls, those in the intervention group reported more satisfaction greater shared decision making, and fewer unmet needs. No change in family stress.	**17**
Havercamp et al. (69)	USA	**Health Profession Education Setting** *Detail:* University	**Health Profession Students** *Detail:* 99 Third-year medical students (50F, 49M) with a mean age of 25.5 years.	**Mixed methods design** *Detail:* Post-test only	**Provider-focused** *Target:* Targeted student comfort and confidence in providing care to autistic children or adults. -----	**Level 1: Reactions** *Detail:* Questions relating to quality of teaching and level of satisfaction with intervention; Reflection that covered the educational experience. Questions on	**Level 1: Reactions-Positive** *Detail:* Within their reflections, students made specific and positive mentions of the	**13**

					Educational intervention for health profession staff/students *Detail:* Online lecture and panel discussion (1 hr-1hr15m)- online lecture covered features of Autism, the value of a medical home, and patient-cantered care. Panel discussion guided to certain topics such as misconceptions, and stigmas associated with Autism; acute healthcare experiences; best and worst experiences with medical professionals.	knowledge about what to do or say during medical exams, and on skills in helping patients feel welcome and comfortable. Reflection that covered the impact of the educational experience on their medical education and future practice. **Level 2A: Learning (Changes in Attitudes)** *Detail:* Questions relating to understanding challenges and confidence and comfort working with autistic patients.	training but also offered suggestions for aspects to improve across the speakers, format, and content; Data were not provided on related questionnaire items. Reflection themes included tools and strategies to use in the future and the importance of communication. A majority of participants reported a positive change on items related to skills in being able to help autistic patients, knowledge about what to do or say when examining an autistic patient and ability to provide better care.	

							Level 2A-Learning (Changes in Attitudes): Positive *Detail:* Reflection themes included better understanding of autism, greater confidence in managing autistic patients, and changed attitudes. A majority of participants reported a positive change in items related to attitudes such as understanding the challenges and confidence and comfort levels.	
Iannuzzi et al. (70)	USA	**Health Profession Education Setting** *Details:* University	**Healthcare Providers** *Detail:* 14 family nurse practitioner students (8 control and 6 intervention)	**Mixed methods design** *Detail:* randomised controlled pilot trial study	**Provider-focused** *Target:* Knowledge about autism, and attitudes towards transition-age autistic youth of nurse practitioner students. -----	**Level 2A: Learning (Changes in Attitudes)** *Detail:* completed the Family Nurse Practitioner Autism Self-Efficacy Scale; and the Societal Attitudes Towards Autism Scale	**Level 2A-Learning (Changes in Attitudes: Mixed** *Detail:* Increase in self-efficacy in working with transition age autistic youths.	**28**

					Educational intervention for health profession staff/students *Description:* Intervention consisted of three components: (1) 3 hours of didactic teaching on autism and best practices; (2) three one hour online modules on epidemiological, medical, and diagnostic issues in autism and information on treatment modalities such as ABA, occupational therapy; and (3) lecture from parents and transition age autistic youths, and 2 hour visit with families of autistic people.	**Level 2B: Learning (Changes in Knowledge/skills)** *Detail:* Autism Knowledge Scale	Societal attitudes towards autism indicative of increased prejudicial attitudes **Level 2B-Learning (Changes in Knowledge/Skills): Positive.** *Detail:* Small increase in knowledge (7.9%) of intervention group as compared to controls.	
Lucarelli et al. (71)	USA	**Secondary Care** *Details:* Hospital outpatient departments	**Healthcare Administrators;** *Detail:* 168 nonclinical administrative personnel, technicians, and clinical nursing assistants from 8 hospital departments	**Quantitative-Group research** *Detail:* Pre/post intervention (immediately post and 1 month delayed post)	**Provider-focused** *Target:* Preparedness of hospital front-line personnel to provide appropriate care for autistic patients ----- **Educational intervention for health profession staff/students**	**Level 1: Reactions** *Details:* survey questions on reactions to the intervention **Level 2A: Learning (Changes in Attitudes)** *Details:* attitudes regarding the importance of Autism-friendly care, comfort	**Level 1-Reactions: Positive** *Details:* 81% of respondents rated the intervention as good or very good, 87% of respondents reported they would be to apply the	**18**

					Description: Following a needs assessment, participants completed an online module with basic Autism knowledge. They then attended in-person training that covered: a case presentation relevant to the department, overview of autism spectrum disorder, video of autistic patient in waiting room and reflection, and strategies treating autistic patients.	responding to the needs of autistic children, and self-reported frequency of behaviours intended to help autistic patients adjust to the hospital setting. **Level 2B: Learning (Changes in Knowledge/Skills)** *Details:* knowledge about hospital resources for assisting autistic children.	training in their current role. **Level 2A- Learning (Changes in Attitudes): Positive** *Details:* Significant improvement on questions related to personnel attitudes and comfort delivering Autism-friendly care and increase in self-reported frequency of behaviours intended to help autistic children adjust to the hospital setting. **Level 2B- Learning (Changes in Knowledge/Skills): Positive** *Details:* Improvement on all questions	

							related to knowledge about autism.	
Mazurek et al. (72)	USA	**Primary Care**	**Healthcare Providers** 14 primary care practitioners (10 paediatricians, 1 family medicine doctor, 2 nurse practitioners, and 1 ‘other’.	**Quantitative-Group research** *Details:* Pre-post intervention (6 months after intervention)	**Provider-focused** *Target:* Addressing the unmet needs for autistic children by training primary care physicians ----- **Educational intervention for health profession staff/students** *Detail:* Extension for community healthcare outcomes (ECHO) autism transition program connecting primary care providers to an interdisciplinary expert team via videoconferencing. Each session consisted of didactic presentations, case-based learning, and collaborative guided practice.	**Level 1: Reactions** *Detail:* Course satisfaction survey. **Level 2A: Learning (Changes in Attitudes)** *Detail*: Self-efficacy questionnaire. **Level 3: Behaviour** *Detail*: Questionnaire evaluating self-reported practice behaviour.	**Level 1-Reactions: positive** *Detail:* Participants reported high satisfaction with the program. **Level 2A-Learning (Changes in Attitudes): Positive** *Detail:* Statistically significant improvements were observed in self-efficacy, **Level 3: Behaviour-Positive** *Detail:* Use of Autism-specific resources.	**22**
Mazurek et al. (73)	USA	**Primary Care**	**Healthcare Providers** 16 Primary care providers (12 completed the program, and 11 completed both evaluations)	**Quantitative-Group research** *Detail:* Pre/post-intervention	**Provider-focused** *Target:* Improve healthcare and reduce unmet needs for autistic children by training primary care physicians ------	**Level 1: Reactions** *Detail:* Course satisfaction survey. **Level 2A: Learning (Changes in Attitudes)** *Detail*: Questionnaire on perceptions of, and barriers to, treating	**Level 1-Reactions: Positive** *Detail:* Participants reported high satisfaction with the program.	**22**

					Educational intervention for health profession staff/students *Description:* An adaption of the extension for community healthcare outcomes (ECHO) autism transition program- 12 weekly one hour sessions connecting primary care providers to an interdisciplinary expert team via videoconferencing. Each session consisted of brief didactic presentations, case-based learning, and collaborative guided practice. Focused on understanding Autism in youth and young adults.	youth and young autistic adults, primary care autism self-efficacy survey **Level 2B: Learning (Changes in Knowledge/Skills)** *Details:* Knowledge test **Level 3: Behaviour** *Detail:* Question about self-reported change in practice	**Level 2A- Learning (Changes in Attitudes): Positives** *Detail:* Significant improvements in self-efficacy, **Level 2B- Learning (Changes in Knowledge/Skills): No clear effect** No significant improvement in knowledge or perceived barriers **Level 3- Bbehaviour: Positive** Most (75%) of participants reported changing their practice as a result of the training	
McGonigle et al. (74)	USA	**Pre-Hospital Care**	**Healthcare Providers**	**Quantitative- Group research**	**Provider-focused** *Target:* To impart knowledge on autism	**Level 2A: Learning (Changes in Attitudes)**	**Level 2A- Learning**	**12**

		Detail: Emergency medical services	*Detail:* 110 EMS personnel and emergency nurses (71M, 39F)	*Detail*: One group Pretest/posttest design	and improve the subjective comfort and awareness of these healthcare providers on how to aid autistic patients in a crisis. ------ **Educational intervention for health profession staff/students** *Detail:* Educational intervention (including training manual and DVD) covering topics such as the nature of autism, myths about autism, medical issues associated with autism, and approach to effectively acquire a history of the acute presentation and conduct a physical examination.	*Detail:* Survey sought information on subjective comfort with responding to acute crises in autism. **Level 2B: Learning (Changes in Knowledge/Skills)** *Detail:* Survey sought information on knowledge of Autism.	**(Changes in Attitudes): Positive** A statistically significant trends post-test for subjective assurance in responding to the acute needs of an autistic patient. **Level 2B-Learning (Changes in Knowledge/Skills): Positive** A statistically significant trends post-test for knowledge acquisition in relation to autism was observed.	
McIntosh et al. (75)	USA	**Health Profession Education Setting** *Details:* University	**Healthcare Profession Students** *Detail:* 27 nursing students in senior year of nursing degree	**Quantitative-Group research** *Detail:* Post intervention only	**Provider-focused** *Target:* Prepare nursing students to care for an autistic patient in crisis ------ **Educational intervention for health profession staff/students**	**Level 1: Reactions** *Details:* 9-item reaction survey.	**Level 1-Reactions: Positive** *Details:* All students agreed with the strategies to teaching, they were enthusiastic about the value	**10**

					Description: Students completed an autism case study in the classroom that provided basic information about autism and for students to apply principles of communication, assessing. prioritising, and critical thinking. Students also participated in a standardised patient simulation and debriefing with a 14 year old autistic teenager who presented to the ED with a laceration in his head.		of the simulation to their practice.	
Mills (76) **STUDENT THESIS/ GREY LITERATURE**	UK (Wales)	**Educational Setting for Autistic Children/Adults** *Detail:* School	**Autistic Children/ Adults** *Detail:* 22 autistic children (12 intervention, and 10 in the control group).	**Quantitative: Group research** *Detail:* Control group comparison, nonrandomised	**Patient-focused** *Target:* Communication with autistic children during medical exams. ------ **Picture Schedules** *Description:* Children in the intervention group were trained to use the ‘show me where’ picture communication system- large posters depicting the back and front of a child with smaller illustrations of	**Level 3: Behaviour** *Details:* Whether the child complied with a physical examination using a single rating scale.	**Level 3- Behaviour: Positive** *Details*: In the intervention group 5 children received a very successful examination, 5 completed with difficulty, 2 partially completed, and 0 were unsuccessful. In the control group of participants	**17**

					different body parts. The children were taught to retrieve appropriate symbols if experiencing pain and comply with examination when presented with a picture.		who did not use the communication system 2 children received a very successful examination, 2 completed with difficulty, 4 partially completed, and 2 were unsuccessful.	
Nicolaidis et al. (77)	USA	**Primary Care**	**Autistic Children/Adults; Healthcare Providers** *Detail:* 170 autistic adults; 41 primary care providers	**Mixed methods design** *Detail:* Pre-post intervention design	**Provider-focused; Patient-focused** *Target:* To make individual accommodations in the provision of healthcare for autistic people ------ **Care Plan** *Description:* Intervention included the autism healthcare accommodation tool (AHAT)- a tool that allows patients to create personalised accommodations report for their primary care provider- and general healthcare and autism related information, worksheets, checklists, and resources for	**Level 1: Reactions** *Detail:* 7 multiple-choice items and 9 open-ended questions about the usability and utility of the toolkit. **Level 2A: Learning (Changes in Attitudes)** *Detail:* Healthcare self-efficacy using a 21 item scale; assessed 16 barriers to healthcare using a checklist; assessed patient-provider communication using an 8 item scale adapted from the 2007 Health Information National Trends Survey (HINTS);	**Level 1- Reactions: Positive** *Detail:* Almost all patient participants (>94%) felt that the AHAT and the toolkit were easy to use, important, and useful. **Level 2A- Learning (Changes in Attitudes): Positive** *Detail:* Comparing pre versus post intervention there was a significant	**24**

					patients and healthcare providers.		decrease in the number of barriers, significant increase in healthcare self-efficacy, and significant improvement in patient-provider communication.	
Riviere et al. (79)	France	**Home and Community; Primary Care** *Detail:* Participants' homes or their medical office	**Autistic Children/Adults** *Detail:* Two autistic children (both M) with autistic disorder (one with co-occurring developmental delays) that were 6 and 8 years old.	**Quantitative-SSRD** *Detail:* Reversal design	**Patient-focused** *Target:* Increased compliance with medical exams (looking in mouth, looking in ears, cutting toenails) ----- **Behavioural intervention drawn from science of ABA** *Detail:* high-p request procedure/ momentum and reinforcement.	**Level 3- Behaviour** Detail: Compliance with a low-probability request.	**Level 3-Behaviour: Positive** *Detail:* Reliable changes in behaviour for both participants by experimental phase.	**22**
Schiff et al. (80)	USA	**Home and Community** *Detail:* Participant's home	**Autistic Children/Adults** *Detail:* One 3 year-old autistic boy with mild developmental delays.	**Quantitative-SSRD** Detail: Reversal design	**Patient-focused** *Target:* Taking liquid medication ----- **Behavioural intervention drawn from science of ABA** *Detail:* stimulus fading and positive reinforcement	**Level 3: Behaviour** Detailed: Correct responding and avoidance behaviour.	**Level 3-Behaviour: Positive** *Detail:* Correct responding increased from 0 to 100% at completion and avoidance went from 100% at baseline to 0% at treatment	**21**

							cessation; Behaviour generalised to performance with his mother in the absence of physician.	
Shabani & Fisher (81)	USA	**Secondary Care** *Detail:* Outpatient clinic	**Autistic Children /Adults** *Detail:* One autistic adult (M) with intellectual disability and type 2 diabetes	**Quantitative-SSRD** Detail: Reversal design	**Patient-focused** *Target:* Treatment of needle phobia to allow for compliance with blood draw procedure ----- **Behavioural Intervention drawn from science of ABA** *Detail:* Graduated exposure and differential reinforcement of other behaviour (DRO)	**Level 3- Behaviour** *Detail:* Percentage of successful trials- non movement of arm during blood draw attempt.	**Level 3- Behaviour: Positive** *Detail:* Clearly differentiated data during the treatment and control phases with no overlap; Improvements maintained at two month follow-up	**13**
Swartz et al. (82)	Canada	**Secondary Care** *Detail:* Pre-admission clinic for Children's hospital	**Autistic Children/Adults** *Detail:* Plans developed for 241 autistic patients who underwent a procedure.	**Quantitative-Group research** *Detail:* post only design	**Patient-focused; Provider-focused; Organisation-focused** *Target:* Intended to optimise perioperative patient cooperation and avoid harm. ----- **Care Plan** *Detail:* An individualised written perioperative plan for autistic children intended to serve as a management guide available to all hospital personnel to optimise perioperative patient	**Level 1: Reactions** *Detail:* caregiver satisfaction measured via a routine postoperative telephone call. **Level 3: Behaviour** *Detail:* Cooperation assessed subjectively within nursing notes and anaesthesia record. **Level 4A: Measures of Results (Changes in Organisation Practice)** *Detail:* use of sedation	**Level 1- Reactions: Positive** *Detail:* 98% of caregivers surveyed were satisfied with the perioperative experience **Level 3- Behaviour: No clear effects** *Detail:* Cooperation at separation and	**17**

					cooperation and avoid harm. Covered factors including optimal time of day for procedure, modification of hospital arrival time, avoidance of multiple transitions, limitation of admission procedures, use of distractors etc.		induction of anaesthesia was 90.2% **Level 4A- Measures of Results (Changes in Organisation Practice): Positive** Cooperation results did not differ between sedated and unsedated patients.	
Whippey et al. (83)	Canada	**Secondary Care** *Detail:* Paediatric hospital- day surgery unit	**Autistic Children/Adults** *Detail:* 18 autistic patients - mean age 8.1 (*SD*=3.5)- from the preoperative clinic scheduled for surgery over 9 month period.	**Mixed methods design** *Details:* Post-intervention only	**Organisation-focused; Provider-focused** *Target:* Experience of aesthetic induction for autistic children. ----- **Care Plan** *Description:* A multidisciplinary care plan was developed for autistic children including environmental modification, and individualised anxiolysis to improve perioperative experience.	**Level 1: Reactions** *Detail:* parental satisfaction, provider satisfaction with aesthetic induction. **Level 4A: Measures of Results (Changes in Organisation Practice)** *Detail:* Assessment of the quality of the aesthetic induction	**Level 1: Reactions- Positive** *Detail:* Parents described the personalised plan, anxiolysis medication, and child life specialist support as advantageous. All nurses, anaesthesiologists, and parents felt the program should continue. **Level 4A- Measures of**	**25**

							Results (Changes in Organisation Practice): Positive *Detail:* 15 (83%) of the aesthetic inductions were described as excellent	
Wolff & Symons (84)	USA	**Educational Setting for Autistic Children/Adults; Primary Care** *Detail:* Group home and primary care clinic	**Autistic Children/Adults** *Detail:* One 41 year-old autistic man	**Quantitative-SSRD** *Detail:* Changing criterion design	**Patient-focused** *Target:* Compliance with needle to skin contact ----- **Behavioural intervention drawn from Science of ABA** *Detail:* graduated exposure and differential reinforcement of alternative behaviour and safety signal (timer)	**Level 3: Behaviour** *Example*: Keeping arm fully on table for duration of needle exposure.	**Level 3: Behaviour-Positive** *Detail:* Participant developed ability to comply with blood draw procedure; Behaviour generalised to primary care clinic and staff there; Behaviour was maintained at 2 week follow up	**19**

Note. SSRD=Single subject research design. * Higher QATSDD Scores are indicative of greater methodological rigour (52). ABA=applied behavioural analysis; IQ=intelligence quotient; QI=quality improvement; EMS=emergency medical services; ED=emergency department

Appendix 4