

Improving outcomes in autism spectrum disorder
through effective service provision: diagnostic
assessment pathways and parent focussed
interventions

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Abstract

Introduction: This thesis on the topic of autism spectrum disorder presents six publications focussed on research about improving outcomes through effective service provision, together with a critical appraisal, which adds depth and breadth to the reasoning and decision making involved in this work. The work follows an iterative process and is positioned from a pragmatist philosophical standpoint, using mixed methods to clarify shared language, concepts and meanings and to ensure translation of research findings into real world practice. The thesis provides evidence of the urgent need for research to inform services as to how they might address the issue of delays in ASD assessment and diagnosis, alongside better understanding of which interventions improve wellbeing outcomes.

Aims: This portfolio of published research has arisen from three related research programmes, with the unifying objectives of:

1. Adding to clinical and research knowledge of current ASD service provision across the lifespan and how that might be improved through adherence to ASD clinical guidelines;
2. Reducing family stress associated with ASD by reducing delays in diagnosis through identification of factors which influence efficiency and quality in a diagnostic pathway, and;
3. Developing understanding of the effectiveness of parent focused interventions to inform future research and practice.

The critical appraisal aims to:

- (a) Explore key areas for debate that have arisen in the work, which transcend the individual publications;
- (b) Position this debate within the context of international literature, research evidence and theory in relation to autism, implementation science and pragmatist epistemology underpinning the work;
- (c) Highlight the contribution of this research to the advancement of clinical practice and research knowledge, and the potential for further clinical reach and informing evidence based practice through diffusion of innovation.

Methods: Research aims, methods and outcomes are presented within a series of publications, using mixed methods to seek to address these.

Findings: Through this research, it was identified that the wait for diagnosis in child and adult services, from referral to diagnosis shared, exceeds the recommended 119 day time standard in 74% of child and 59% of adult services. There was a significant difference in mean age of referral and diagnosis for girls compared to boys and this delay occurred prior to referral through delayed recognition rather than through delays in the assessment process. Findings provide strong evidence of the need to address the way we collectively deliver ASD services.

This research identified factors which influence waiting times in child and adult services. These included the availability of relevant pre-referral information at first appointment, consideration of the number of contacts or appointments used to reach a conclusion and complexity of the case. Mixed methods were used to further identify a broader range of factors affecting wait times in each service and to develop child and adult action plans as proposed solutions that could be applied by local service providers. Plans developed were then successfully applied in a 12 month service improvement intervention with 11 adult services, resulting in a statistically significant reduction in duration of assessment ($b=-0.25$, $t(136) = -2.88$, $p=0.005$), taking the duration to within the recommended timescale. In child services, this model also led to a statistically significant reduction in waiting times for diagnostic assessment and increased identification of girls with ASD.

Systematic review and meta analysis of parent focused intervention for older children and adults found that a) parent training and education and b) mindfulness interventions provide measureable improvements in family wellbeing, which in turn are known to have a reciprocal effect on the individuals with ASD. A number of recommendations for future research arise from this work. These include the aspiration for an ASD specific wellbeing measure which can be applied with greater consistency across ASD intervention studies; and the need for consensus on theoretical models to underpin evaluation of complex interventions in ASD, which in turn may lead to deeper understanding of which elements of interventions are most effective in which circumstances.

Conclusion: In a field where research evidence has been lacking, this body of work applies a range of research methods, in order to add to the evidence base and provide practical steps, which clinical service providers could apply, to reduce the delays in diagnosis. Earlier access to ASD specific interventions, as a result of earlier diagnosis, has the potential to improve wellbeing for individuals with ASD and their families. Despite limitations of the research on ASD parent focused interventions for older children and adults, findings reported here add further support to the ASD clinical guideline recommendations to use such interventions.

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Preface

The critical appraisal that follows, together with a selection of my publications, is the culmination of six years of research, preceded by over 20 years of clinical experience as a speech and language therapist in the field of Autism Spectrum Disorder (ASD). My research focus has been closely related to my clinical practice and concentrates on seeking to continue to extend a broad and deep knowledge of ASD service provision for children and adults through the development and application of evidence.

The publications demonstrate the use of mixed methods to unite and make connections between related fields of enquiry from speech and language therapy, occupational therapy, education, psychology, practice development, and implementation science, which reciprocally seek to improve the support offered to individuals with ASD and their families.

The papers selected represent three linked research programmes, each grounded in the desire to make changes in practice.

Six publications (referred to as papers 1 to 6 throughout) are presented as representative of this interconnected body of work. The appendix includes abstracts from each publication, and co-author statements from two co-authors to provide evidence alongside my own self evaluation of my personal contribution to this published work.

These papers are not an exhaustive list of my output on this topic and additional papers are referenced in the traditional manner and outlined in my curriculum vitae in the appendix.

Papers are presented in their research programme, starting with research programme one, summarised below.

Research Programme 1:

What is the wait for diagnostic assessment in Scotland; which factors influence delays and what can be done to reduce the wait?

- **Papers 1-3:** Papers 1 and 2 present the quantitative results from the research on describing the problem of delays in diagnosis in children and adults with ASD in Scotland, and the factors which influence this delay. Paper 3 presents key qualitative results. Data from focus groups with clinicians were triangulated with quantitative data and synthesised into co-produced action plans for child and adult services. These plans were then used to guide the reduction of delays in diagnosis, with real world application.

Research Programme 2:

Impact: Can an ASD focused practice development model be used to reduce the wait for diagnostic assessment in child and adult clinical services?

- **Papers 4-5:** Following Research Programme 1, it was important to understand whether this evidence could be applied within practice and how effective this would be. Although the wait was longer for child services, there continued to be a significant lack of evidence about ASD in adults and therefore this was selected as the initial focus of research in research programme 2. Paper 4 focussed on evidence reported from a programme of work to apply evidence from programme one, in a 12 month practice development implementation programme in 11 adult diagnostic services. In my dual roles of practitioner and researcher, I was also motivated to bring these worlds together and to lead a similar programme of change within my own clinical service, which was also focussed on addressing waiting times. This approach was found to be effective in improving waiting times for assessment in child services over a 2 year period of data collection (Paper 5). Papers 4 and 5 provide evidence of the effectiveness of the model of service improvement reported and applied across research programmes 1 and 2. It is well known that making real world impact with research is challenging; papers 4 and 5 reporting success in reducing the delays in ASD diagnostic assessment complete the body of work whilst setting the stage for further development of my research.

Research Programme 3:

Do parent focused interventions for parents of older children and adults with ASD improve wellbeing outcomes for parents?

- **Paper 6:** There is no single intervention recommended for all people with autism, however clinical guidelines have recommended a small number of approaches for which there is sufficient evidence. One of these is psychosocial interventions focussed on parents of adults or children with ASD. Despite NICE and SIGN clinical guideline recommendations that parent focussed interventions are offered across the lifespan, this decision was made largely based on extrapolating evidence for the effectiveness of parent mediated interventions for younger children. There is in fact, limited evidence about which specific interventions or approaches to use for parents of children over 7 and adults. Additionally, there is a lack of consensus on which outcome measures should be applied for interventions targeting parent wellbeing, through increased knowledge and skills to enable them to support their child. In paper 6, I undertook a systematic review and meta analysis, to consider this question, providing an overview of the literature and current evidence about parent-focussed interventions in ASD. This paper is submitted and is under second review at the time of submission.

Selected publications

Paper 1

Rutherford, M., McKenzie, K., Johnson, T., Catchpole, C., O'Hare, A., McClure, I., ... & Murray, A. (2016a). Gender ratio in a clinical population sample, age of diagnosis and duration of assessment in children and adults with autism spectrum disorder. *Autism*, 20(5), 628-634.

Paper 2

McKenzie, K., Forsyth, K., O'Hare, A., McClure, I., Rutherford, M., Murray, A., & Irvine, L. (2015). Factors influencing waiting times for diagnosis of Autism Spectrum Disorder in children and adults. *Research in developmental disabilities*, 45, 300-306.

Paper 3

Rutherford, M., McKenzie, K., Forsyth, K., McCartney, D., O'Hare, A., McClure, I., & Irvine, L. (2016c). Why are they waiting? Exploring professional perspectives and developing solutions to delayed diagnosis of autism spectrum disorder in adults and children. *Research in Autism Spectrum Disorders*, 31, 53-65.

Paper 4

Rutherford, M., Forsyth, K., McKenzie, K., McClure, I., Murray, A., McCartney, D., ... & O'Hare, A. (2018a). Implementation of a practice development model to reduce the wait for Autism Spectrum diagnosis in adults. *Journal of autism and developmental disorders*, 1-15.

Paper 5

Rutherford, M., Burns, M., Gray, D., Bremner, L., Clegg, S., Russell, L., ... & O'Hare, A. (2018b). Improving Efficiency and Quality of the Children's ASD Diagnostic Pathway: Lessons Learned from Practice. *Journal of autism and developmental disorders*, 48(5), 1579-1595.

Paper 6

Rutherford, M., Singh-Roy, A., Rush, R., McCartney, D., O'Hare, A., & Forsyth, K. (2019). Parent focused interventions for older children or adults with ASD and parent wellbeing outcomes: A systematic review with meta-analysis. *Research in Autism Spectrum Disorders*, 68, 101450.

Critical appraisal

Improving outcomes in autism spectrum disorder through effective service provision: diagnostic pathways and parent focused interventions

Introduction

Internationally, there is little doubt about the need to improve outcomes for autistic individuals and their families through effective service provision (Howlin et al., 2000; Steinhausen et al., 2016), nor about the need to make decisions about improvements based on best available research and practice based evidence (SIGN, 2016). There is less consensus on how outcomes are defined (Henninger & Taylor 2013) or on how to improve outcomes in ways that are contextually relevant to the autistic individual across the lifespan, their family, practitioners and services delivering support, policy makers and legislators.

In real world research, and specifically in this work, questions and proposals originate from the need to answer a practical question, rather than from philosophical affiliation or theoretical drivers. This is very much reflected in the style of writing in the publications submitted. However, doctoral study affords an opportunity for deeper reflection on the philosophy and theory underpinning the practical work and its dissemination.

In this critical appraisal I will explore some of the central concepts and assumptions underpinning my approach to the study of improving outcomes for autistic people and the decision to focus on diagnostic pathways and parent focused interventions. I will reflect upon the steps taken to support diffusion of innovation and relevance of research to practitioners and autistic people together with the strengths and limitations of the approach applied.

Aims

This portfolio of six published research papers has arisen from related research programmes, with the unifying objectives of:

1. Adding to clinical and research knowledge of current ASD service provision across the lifespan and how that might be improved through adherence to ASD clinical guidelines;
2. Reducing family stress associated with ASD by reducing delays in diagnosis through identification of factors which influence efficiency and quality in a diagnostic pathway, and;

3. Developing understanding of the effectiveness of parent focused interventions to inform future research and practice.

The critical appraisal of this portfolio aims to:

- (a) Explore key areas for debate that have arisen in the work, which transcend the individual publications
- (b) Position this debate within the context of international literature, research evidence and theory in relation to autism, implementation science and epistemology underpinning the work
- (c) Highlight the contribution of my own research to the advancement of clinical practice and research knowledge, and the potential for further clinical reach and informing evidence based practice through diffusion of innovation.

Methods

Each publication (papers 1-6) includes a methods section for that study, however, the critical appraisal provides an opportunity for more in-depth critical consideration of epistemology, methodology, areas of key philosophical debate in ASD research and decision making around theories in implementation science. In the systematic review and meta-analysis, the issues that arise for critique include placing this research within the context of related research and more in depth discussion about interpreting findings.

The mixed methods research reported draws on theoretical and philosophical perspectives, which are grounded in pragmatism. This is a philosophical position, which places knowledge in context (Creswell & Plano-Clark 2011) and tests knowledge through empirical enquiry in a manner, which allows a more complete story to be told (Morgan 2007). Regardless of philosophy, all researchers endeavour to find the truth and reflect reality through authenticity of approach. Within a pragmatist paradigm, truth, reality, evidence and knowledge are strongly intertwined with their usefulness and practical application as well as their ‘meaningfulness’ to those they impact upon.

Pragmatism

Historically the very nature of the opposing stances of constructivists and positivists or post positivists left the researcher in the position of making a choice between them, with qualitative and quantitative researchers on two opposing sides viewing “truth” as either an objective

reality or a subjective construct (Tashakkori & Teddlie 2010). However, the pragmatist stance represents the enticing possibility of a shift away from more black and white perspectives, to those, which consider the overlaps and benefits of applying different theories because they make practical sense in any given context. The “truth” from the pragmatist perspective is present in a shared understanding of a phenomenon under study, which may come from both objective and subjective measures. Shared actions and practice which emerge from this shared view of knowledge, contribute to the attractiveness of this approach to the practitioner-researcher determined to make links between “the truth” and evidence base from research and the useful application of this in the real world (Denscombe 2010).

One could argue that constructivist and interpretivist frameworks provide equally valid positions from which to consider this work because they place knowledge in context. They also support the use of mixed methods in gathering information in the most appropriate way to the question being posed, with an emphasis on practical solutions (Cresswell, 2014). However having reviewed a range of epistemological paradigms, I have returned to my roots as speech and language therapist. I am drawn to the pragmatists because of their focus on communication, making shared meaning and narratives, which represent meaning through language, societal norms and expectations. For pragmatists, intersubjectivity and transferability are central (Shannon-Baker, 2016) and in this work involving practitioners, these are demonstrated in the design of the studies, which engage practitioners in dialogue. The purpose of this dialogue is to identify shared language and meaning around the constructs of focus, to measure and improve waiting times through practical solutions generated, based on evidence gathered from their local area.

Effective dissemination is central to meaningful research and this complex and many layered process, starts with a question that is relevant and useful to a particular community. In order to meet the aims of this research, an academic-practice collaboration was formed to engage with relevant communities in the design and delivery of the research. The intention behind this decision was to approach the research with an understanding of a broad range of perspectives on the questions to be addressed, rather than a single lens. The initial focus on waiting for diagnosis was initiated by the Scottish Government and was relevant to the research- academic team who included members involved in diagnostic assessment teams across the lifespan and developmental stages. This team had experience of trying to manage demand and capacity within clinical services and regularly engaged with the autism community in terms of their preferences and challenges in receiving timely and appropriate support. The issue is also well

reported in research with autistic people and their families, and more specifically, it arose from the autistic community in Scotland, as reported in the Scottish Autism Strategy in 2011.

Assumptions and concepts within autism research

Within a pragmatist framework, the researcher is interested in the meaningfulness of the research for those it impacts upon. Papers 1-3 provide ‘objective’ information and evidence about wait times for autism assessment and factors, which were found to influence this. Participants in implementation focussed research reported in papers 4-5 were practitioners who may or may not have had access to this information prior to their involvement in changes to their own local pathway for assessment. They may or may not believe this information to be important or true. These variables would affect their own motivation and whether they do or do not expect to change their own practice. There is therefore a range of assumptions and concepts it was important consider in developing shared language and shared understanding of the focus of the research, in order to engage practitioners in practice change to reduce the wait for autism assessment and diagnosis, which are explored below.

Practitioner knowledge, beliefs and perspectives underpinning the adoption of new roles in practice

Within the pragmatist framework, the concept of waiting for ASD diagnosis is defined in the doing rather than the being (Benton and Craib, 2011), so that the “end-users” (Carpenter et al., 2005) of the evidence were practitioners and services delivering ASD diagnostic assessment. For services to participate and adopt a new model based on the research, it should resonate with their values, beliefs and current needs of the organisation (Dingfelder & Mandell 2011). This would require acceptance of the strong evidence that the wait for ASD diagnosis was too long and required reduction (paper 2). There needed to be agreement over the position that an earlier ASD diagnosis was not only cost effective (because services are more efficient and fewer appointments are needed to reach the same outcome) but also that this was beneficial because it reduced stress and supported earlier access to appropriate services. In turn, this can reduce the escalation of more challenging behaviours and experiences. The model proposed set out an expectation of a) increasing the number of ‘generalist’ practitioners engaged in ASD diagnostic assessment for less complex cases and b) that with support, practitioners and researchers in leadership roles would take on new roles, with additional training but without increased resource. It was important that service providers perceived the model of service delivery as practical and realistic.

Although the ‘autism experts’ within the participating services were ‘self-selecting’ and motivated to improve diagnostic services, there was a need to revisit some assumptions and philosophical positions with ‘generalist’ practitioners, who previously worked with and assessed many individuals with ASD but who did not personally make the diagnosis, such as ‘is diagnosis helpful at all?’ and ‘is quicker diagnosis better?’.

Is diagnosis helpful?

One assumption worthy of debate was whether or not the practice of giving a diagnosis or a label is in the best interests of people meeting criteria and what are the pros and cons of this. This is a debate likely to continue, amongst people with fairly polarised views of what is conceived of as a ‘medical model’ or a ‘social model’ of disability (Graham and Tancredi, 2019). From the pragmatist position, both of these have relevance when we consider context, leading me to conclude that my work is framed by the view that diagnosis can enhance understanding of support needs and access to relevant support within our current society but support should not wait for diagnosis.

I was able to reflect and reach consensus with colleagues, that although there are always exceptions to the rule, we should take the position that we will use diagnostic labels for reasons outlined below.

In order to carry out meaningful research, the use of international diagnostic classification systems supports clear definition of the research sample and results that are transferable to other equivalent populations (SIGN, 2016; NICE, 2011). Evidence indicates that diagnosis leads to access to more appropriate and timely support (Shattuck et al., 2009). Further evidence suggests that the diagnoses made are reliable and stable (Moore & Goodson, 2003; SIGN, 2007; NICE, 2011) and consequently, the results of intervention studies can be generalised to others with the same profile and diagnosis. ASD diagnostic criteria describe a spectrum of presentations, thereby highlighting the need for individualisation of supports and recovery focussed services (Slade, 2009) emphasising the potential for change and progress. Beyond the academic evidence, many organisations representing autistic people and their families are supportive of using diagnostic labels and they campaign for better access to early and timely diagnosis for children and adults (e.g. National Autistic Society).

For all of these reasons, I believe that the practice of using the ASD diagnostic label is of more overall benefit than harm. However, as a reflective practitioner and researcher, I must also consider opposing positions and be open to continued debate.

Others would argue that labelling inhibits individuals because it creates stigma and a stereotyped view rather than an individualised understanding of the person (Anastasiou & Kauffman, 2011). Prejudice in society may negatively affect the participation and opportunities of those who openly disclose their “label” (Corrigan & O’Shaughnessy, 2007). There is a juxtaposition between the challenges of identifying those who need a particular range of supports, so that they can access these in a timely way, against the reality of the uninformed majority or even diagnosed individuals themselves who may naively set limitations, discriminate and make assumptions that the person with ASD cannot change. On the other hand, whether there is a diagnosis or not, the presence of the challenges of ASD can often lead to social exclusion (Howlin, 2013). Therefore, I have argued that, as well as supporting the individual to understand why aspects of life have been difficult, the diagnosis provides a short hand to those around the person, to enable them to enable the individual with ASD. It also guides an evidence based approach to practice (SIGN, 2016). Whilst I acknowledge the important aspiration that diagnosis should not be a requirement for support (Tisdall, 2015), the reality is that diagnosis can facilitate better support. I currently continue to believe that there is a core challenge, which can reliably be identified as the multi-dimensional condition of ASD, and that on balance, labelling (where it applies) is currently likely to be beneficial. Equally, each case must be considered in its individual merit and there may be exceptions to the rule. For colleagues wrestling with such a question for an individual, access to peer review, support and mentorship would be explicitly offered in the pathway implemented.

In this work, knowledge tested through empirical enquiry is viewed as intertwined with its practical application and therefore not always ‘black and white’ (Morgan, 2007; Morgan, 2014; James, 2014). There will always be a subjective element to ASD assessment and diagnosis, which I consider to be as much of an art as a science and I hope that through broad and deep consideration of the range of positions amongst the autism community, the quality of the research is strengthened.

Is quicker diagnosis better?

In our culture and social organisation in Scotland, we have an NHS, in which waiting times are used as an important, if controversial measure of success of service provision (Morton & Bevan, 2012; Barbour et al., 2014). Although the concept of an optimal duration for diagnostic assessment is affected by local social constructs, this is an issue with relevance in the UK and European context (NICE, 2011; Davidson, 2014) as well as internationally, for example in Canada (Penner et al., 2018), Australia (Whitehouse et al., 2018), the USA (Gordon-Lipkin et

al., 2016) and India (Barua et al., 2017). One assumption within our programme, which was questioned by participating service providers, is whether shorter waiting times, quicker assessment duration and earlier diagnosis are in fact desirable, particularly for parents of very young children.

Research is improved when it takes account of the perspectives of those the research impacts upon (Heikkinen et al., 2012; Shickle et al., 2014). I was therefore; keen to listen when participants highlighted that optimal duration of diagnostic assessment might not be the fastest duration. This was despite research evidence to support my own position (Sansosti et al., 2012; Martinez et al., 2018) with many examples of evidence of the reported benefit to knowing what the “problem” is and that diagnosis can facilitate access to appropriate interventions (Shattuck et al., 2009). My understanding was that long diagnostic delays can lead to a loss of confidence in healthcare professionals, and that families are more likely to seek alternative treatments for their child, which have poor empirical support (Harrington, et al., 2006).

However, through clinical experience I understand there are many factors involved in making a diagnosis, which may require patience and time appropriate to each individual. These include setting and meeting expectations, which are communicated effectively to the individual and family. Some families may need time to process and understand the assessment and diagnostic process in relation to a neurodevelopmental and lifelong condition, whose presentation is heavily related to external as well as internal factors.

Therefore, there is philosophical merit in the view that extending the process of diagnosis unnecessarily is to be avoided but that a degree of individualisation is also necessary and that diagnosis is not the ‘be-all and end-all’ in any ‘clinician-patient’ partnership. In addressing this concern, participants and researchers were able to reach agreement that professional judgement was required in each case and that there would be an open channel for discussing cases as they arise. As a result, we have reached a more nuanced consensus position on the way the pathway and wait time standards are applied.

Understanding efficiency in services and measuring the delay

Delayed diagnosis is commonly reported as a source of stress for families and dissatisfaction with services (Moh & Magiati., 2012; Crane et al., 2015). If we accept that making diagnosis quicker is a valid aim, then defining how wait times or ‘efficiency’ in a service would be measured was therefore an important first step. I took the decision to refer to the only existing published measurement example - the United Kingdom (UK) National Autism Plan for Children (Le Couteur et al., 2003). This guidance recommended that the timescale for

diagnostic assessment of children is no longer than 119 days (17 weeks) from referral for ASD assessment to diagnosis being shared (Le Couteur et al., 2003), however, only some child services meet this standard (Palmer, 2011) and many others are unable to report on their wait times at all. At the point of embarking on this research, there was no report available to evidence the wait times across Scotland and no clarity over the extent of the problem. This research made explicit reference to information to be gathered at each stage in the pathway, including the number of clinical contacts, types of assessment undertaken, key time points (date of referral, date of first assessment, date diagnosis made and date diagnosis shared) and the time between these. There is no recommended wait time standard for adult diagnostic services in the UK (or elsewhere) and therefore I applied the child standard for comparison in this research, which is close to the NHS mental health standard for wait time to first assessment of 18 weeks.

In order to build in relevance to the autism community and support dissemination, the data collection tool was built with reference to the above and through consultation with an external international expert reference group of researchers and practitioners (Paper 2) and the information about how the delay would be measured and why, was shared with participants.

Measuring Quality

Within a pragmatist paradigm, where meaning is defined in the context in which it is relevant, efficiency is clearly an important element of ASD clinical services, battling with long waiting lists and increasing referral rates. Resources are finite and their efficient deployment is essential to high quality services. However, efficiency should never be at the expense of the robustness and quality of the assessment process. Evidence based practice is expected, as demonstrated through adherence to quality standards or clinical guidelines for diagnosis and assessment (NICE, 2011; 2012; SIGN, 2007; 2016). Busy clinicians do not always have time to reflect upon aspects of quality within their service and there can be differences of opinion about interpreting clinical guidelines and whether to aim to apply a ‘gold standard’ or a ‘good enough’ standard of autism assessment and diagnosis (Matson et al., 2012).

Evaluation of the implementation of clinical guidelines in practice is an important component for their further improvement (Nivoli et al., 2011) and conversely audit of adherence to clinical guidelines is an important factor in delivering quality services, applied across many clinical areas (Hysong et al., 2006). There are many critical reviews of clinical guidelines (Pearson & Rawlins, 2005), which highlight that “guidelines can work but often don’t” (Ltd, 2005, p274) because lack of practitioner adherence. As a pragmatist, I am interested in why this is and how

we can address the issue. Adherence is affected by reduced awareness of the existence of the guidelines; limited consensus about how to interpret them; insufficient training and guidelines perceived as impractical or requiring additional time (Rhodes et al., 2010). The time lag between publication of evidence for effective interventions or standards and implementation of these in practice is commonly reported (Dingfelder & Mandell 2011; Grimshaw et al., 2012).

In order to address these concerns, this research includes a measure of adherence to quality standards in the data gathering tool (reported in paper 2). We undertook further analysis of whether there was a relationship between greater adherence to guidelines and waiting times (McKenzie et al., 2016a; McKenzie et al., 2016b) and fed this data back to participants (paper 3). The opportunity for participants in the research to reflect upon data on quality in their own service, was built in to the mixed methods approach. This was intended to inform the development of local actions to improve services, which reflect aspects of quality assessment linked to clinical guidelines and which were important to participants. The explicit focus on intersubjectivity was intended to build mutual consensus on what constitutes a good quality service and to support the generalisability of the research through implementation.

Complexity

ASD is a complex condition because of the heterogeneity of aetiology and behavioural presentation (Damiano et al., 2014) and because co-morbid conditions are present in most cases (Mazefsky et al., 2012). NICE (2012) references the concept of complexity in autism assessment, suggesting that standardised tools are useful in more complex cases but this in itself is a notion requiring further definition.

Philosophically, I come from the position that environment or context is as important as individual factors in determining how ‘complex’ the assessment of an individual for autism might be. However, historically complexity has been defined as a ‘within – patient’ construct and therefore reaching ‘shared understanding’ about complexity and how it affects effective and efficient models of service provision would be important to support dissemination and implementation of the research.

Complexity of a “case” can arise on the one hand in relation to the individual being assessed – for example, where individuals have a presentation in keeping with more than one co-morbid diagnosis such as genetic or chromosomal abnormalities; ADHD; attachment disorder or learning disability (Damiano et al., 2014). Individuals may be very poorly regulated and hard to engage in assessment. For subgroups where diagnosis is commonly delayed, there may be

complexity in their presentation (e.g. girls, looked after children or children from bilingual families) (NICE, 2012). For other individuals, there may be discrepant findings between reported and observed ASD signs or signs across contexts. Although this is not mentioned in research or guidelines that I have found, it is my view that complexity can equally result from factors relating to the skill, experience and resources of the clinical team, so that if the team usually sees school aged children, they may view a pre-school child as complex, whereas another team may not. Finally, there are factors relating to the individual's environment, such as possible missing information, affected by the level of access to a person who can give a good quality developmental history; emotions or attitudes towards the diagnosis of ASD within the family or community or indeed the suitability of support provided to the individual (paper 3). Individuals, who have their needs well met, may present with less obvious ASD signs and others can 'mask' or 'camouflage their challenges in attempts to 'fit in' (Hull et al., 2017) all of which can add complexity.

In the data collection tool, and in resources developed for pathway implementation, key factors affecting complexity were included in order to determine whether services and practitioners made different clinical decisions where aspects of complexity are present and whether complexity affects wait times. Through the inclusion of this data, the next phase of research could consider which improvements might be made based on this knowledge. This construct also then formed the basis of the 'effective and efficient' pathway model which leads the team to allocate referrals to an 'abbreviated' pathway or a 'complex' pathway.

Gender: Women and Girls

There are certain demographic factors and individual, familial and environmental risk factors which do seem to increase the risk of delayed diagnosis and in turn reduce the opportunity to access ASD specific interventions. (Daniels & Mandell, 2014; Rutherford et al., 2016a). I made the decision to focus in more depth on the very topical factor of female gender, in order to improve understanding about how this variable affects different stages of the pathway (referral, assessment duration and diagnosis). Autistic girls and women are known to be particularly disadvantaged through under-recognition, misdiagnosis or delayed diagnosis (Giarelli et al., 2010; Shattuck et al., 2009) as outlined more fully in paper 1. It is likely that there is a societal perspective on gender, which also impacts on autism diagnosis, so that parents and clinicians judge and report the same behaviours differently in boys and girls. Given that the diagnostic criteria applied are the same regardless of gender, this study could gather data to examine whether there is a delay in diagnoses for females in Scotland, although

research methods applied would not gather data about whether this was an appropriate delay because of a specific reason.

Prior to this research there was little evidence to clarify at which point in the pathway this 'delay' arises and looking at this variable would allow us to consider whether it is a lack of skill, knowledge or experience in referring and/ or diagnosing teams in relation to different presentations of autism.

Data gathering protocols were designed to ensure that evidence related to gender were collected, in order that this could be included in the implementation phase based on evidence rather than speculation.

Adults

There is also very limited research evidence about adults with autism (NICE, 2012) and therefore this work provided an opportunity to include this population. The question of diagnostic 'delay' is further complicated by the fact that autism is a neurodevelopmental condition, present from childhood, so that any diagnosis in adulthood could be considered to be delayed. Service provision is very different than for children, therefore it was important to ensure data collected was analysed and reported separately for adults and children. The mixed methods approach would allow consideration of commonalities in challenges and solutions across the age range.

Mixed methods

The use of mixed methods, creates a requirement for the researchers to have equally strong rigour and reasoning for both quantitative and qualitative methods applied. Through a sequential mixed methods design (papers 2 and 3), quantitative data were collected from retrospective case note analysis of a stratified randomised sample, in order to use recent 'real world' cases to collect nationally representative data on waiting times (paper 2). Proportionate stratified sampling was undertaken through randomising a sample frame of all services offering diagnostic assessment in Scotland. This approach was used to infer that the sample was representative of the Scottish population and to support generalisability of results. Power analysis was applied (Stapleton, 2006), to determine the ideal sample size for regression analysis of data collected to determine which factors influenced wait times. Power is the probability of detecting an effect, assuming the effect is really there and the calculation aims to ensure sufficient sample size to detect an effect. The recommended magnitude of effect size is mathematically calculated and depends on the effect being considered. In this case a medium

effect size of 0.3 was agreed, which in turn affected the number of data points that could be included in the analysis. We knew this from the outset and ensured key variables could be included, through use of initial bivariate correlations between all sociodemographic factors, ASD risk factors and the three outcome variables. Five important predictor variables were identified, with number of contacts as the mediator variable. Data were analysed through regression analysis, because this allowed comparison an independent variable with more than two predictors and to look both at the effect size and the direction of the effect. As well as answering the research questions in relation to wait times, they were fed back to practitioners who provided the data.

Qualitative methods were then applied to use this data meaningfully and to further explore reasons for waits for diagnosis and practical solutions developed through focus groups with practitioners. In this way the mixed methods approach and pragmatist stance, led to shared 'truth' and 'meaning' about not only what would improve wait times, but how this could be done practically within current service provision and resources. The ideas and 'action plans' developed were locally relevant and based on quantitative evidence, as well as practitioner narratives and ideas from experience. Adult and child services could create solutions relevant to needs of their populations (paper 3). There is the potential for generalisability because of a) the sampling method to identify a nationally representative sample; b) commonalities in the issues under discussion and c) in the shared context of a nationalised healthcare system.

In qualitative research, demonstrating rigour and power of a study is just as important as it is in quantitative research and this was considered in the design. There is no evidence based guide to sample size for qualitative research, although a sample of 95 professionals, from 16 services for the focus groups is a relatively large number. The number is less important than the judgement about whether this is indeed a big enough sample to generate an adequate breadth and quality of responses to the focus questions. Key considerations were that the work was undertaken with practitioners rather than 'patients'. Practitioners are likely to be less disempowered, more articulate and capable people who can engage in quality dialogue. They hold key 'information power' and 'narrative coherence' important in considering sample size and methods in qualitative research (Malterud et al., 2016). The study applied a purposive sample through stratified randomised sampling to permit cross service thematic analysis from data gathered from 16 different services. In order to increase the power of information available, reduce the need for a larger sample size and to therefore increase generalisability, there was a relatively narrow study aim and a structured framework for data gathering through focus groups. Participants were asked to focus on very specific questions related to objective

data on their own service, to support the development of a shared understanding of the key issues.

Shared actions and practice, which emerge from a shared view of knowledge, contribute to the attractiveness of the mixed method approach to the practitioner-researcher with a determination to make links between “the truth” and evidence base from research and the useful application of this in the real world (Denscombe 2010).

The application of mixed methods added depth and richness to research findings because we did not only seek to describe the “problem” but also to understand it (Cresswell, 2014) and to actively take steps to engage practitioners with ways to apply the evidence in practice. Within an action research framework from the pragmatist philosophical perspective of seeking to place research within a meaningful real world context, describing the problem and then identifying potential solutions, were only the first steps. The obvious next step was to seek implement the recommended “action plans” with services. This required the application of research methods and theory drawn from implementation science. Practice development theory and implementation science, as discussed below; sit well within a pragmatist position, to facilitate the diffusion of innovation (Dingfelder & Mandell 2011). I would argue that the research described in papers 4 and 5 refers to complex ‘real world’ interventions, focussed on many practitioners across services, making a range of adaptations and changes to practice relevant to their local context, based on both research evidence and practitioner skill and experience. I would therefore like to give further consideration and critique of research methods, theoretical frameworks and underlying assumptions, which form a basis for planning a novel service change intervention in the context of a complex multi-disciplinary system, within a pragmatist framework.

Implementation Science

It takes time for new interventions to be adopted or implemented in public services (Dingfelder & Mandell, 2011) and the universal research – practice gap (Grimshaw et al., 2012) applies within the field of ASD. This is in part because knowing what needs to change does not equate to practice change with meaningful impact (Melton et al., 2010) and it is known that targeted implementation programmes, which are informed by theory, can accelerate change, more than a ‘train, inform and hope’ attitude.

Implementation science is the study of approaches to understanding and evaluating the uptake of evidence and research into ‘real world’ practice (Douglas & Burshnic 2019). This is very much in keeping with a pragmatist epistemology, where knowledge and evidence are

understood through consideration of the perspectives of those involved and through contextual relevance. Implementation research applied to autism health service provision, requires a broad-based epistemological approach, which combines a range of sources of information (McCurrtin et al., 2019). Although still a relatively new academic field at the time of my own research in 2012 (papers 4 and 5), in recent years there has been a “rapid accumulation, systematisation, and advancement of knowledge about implementation strategies, actors and contexts” (Kislov et al., 2019). This is therefore, a topic worthy of further academic consideration beyond the results reported in the 6 published papers submitted.

In research with a basis in implementation science, methods are applied which consider that services using the evidence are ‘complex systems’ influenced by a range of individual and environmental factors. There is a focus on the processes, mechanisms influencing change and outcome(s) of whether or not the new practice was adopted rather than (or together with) a focus on intervention outcomes (Douglas & Burshnic 2019).

In order to continue to build on prior research recommendations and put them into action, the research in paper 4 applied knowledge derived from implementation science. The outcome of focus was on whether or not it would be possible to reduce the wait for assessment and diagnosis. This was targeted through two combined methods of a) applying a practice development framework, which takes account of mechanisms of practice change, and b) applying processes identified in papers 1-3 and summarised in the Autism Achieve Alliance action plans and pathways and proformas (AAA, 2013). Our prior research did not seek to identify mechanisms to support practice change and therefore we sought and identified a published framework to explicitly take account of known mechanisms. This framework, named ‘Flightgate’ draws on ‘practice development theory’ (Melton et al., 2010; Melton et al., 2012) to propose key mechanisms, which influence practitioners in implementing practice, change.

Theories of practice development

A range of implementation frameworks have been applied in healthcare, outlining key steps required in the complex, dynamic process of service change (Meyers et al., 2012) and offering a conceptual structure to explain and predict why an implementation programme is successful or not (Harvey & Kitson, 2015).

Philosophically, these sit well within the pragmatist paradigm, which continues to be a relevant position from which to apply the research methods in papers 4 and 5. In this work, the aim is to recognise the complexity in the real world contexts and methods applied to implement

multifaceted solutions nationally in a range of services, each with their own complex configuration of service culture and individual practitioner perspectives. Practice development frameworks recognise not only the importance of context but also the importance of the interactions between a range of complex individual, interactional and environmental factors which can be mechanisms for change (Pfadenhauer et al., 2017).

Such practice development frameworks have been widely adopted in the nursing profession (McCormack et al., 2013), with more limited application amongst allied health professionals (Bradd et al., 2017) or within autism services (Drahota et al., 2012). At the time of planning our own research there was no model or framework specifically recommended for service change in multi-disciplinary autism diagnostic services.

As the field of implementation science has developed, there has been some criticism within the medical community, of slow progress hindered by a lack of specification of implementation strategies for different areas of clinical practice or mismatch between implementation strategies and the ‘problem’ being targeted (Wensing & Grol, 2019). A further criticism is the limitations in the availability of statistical methods to test the direct or indirect mediating and moderating effects of mechanisms being measured and a lack of standardised measures to link mechanisms to outcomes (Williams, 2016; Beidas et al., 2019). Studies are criticised for not including end-user feedback into the implementation intervention design.

With hindsight, it is important to reflect upon our own decision making over the selection of the ‘Flightgate’ framework based on practice development theory (explained fully in paper 4). I can see that some of the above criticisms of a field in its infancy equally apply to our research, which sought to apply evidence based implementation strategies but did not seek to explicitly measure the individual and contextual factors, which may have influenced the outcome of reduced waits for assessment and diagnosis.

Within the limited timescale of a 12 month real world research programme, I searched the literature for examples of other models of change and frameworks used in autism services but did not find published examples. Although, I then searched more broadly for implementation models applied in healthcare, I did not find any that stood out as having a greater justification for application in this work than the Flightgate framework. This published framework identified that different types of ‘Practice Development’ are more or less effective for different individuals in implementing focussed changes to practice, depending on contextual factors, which can be personal or environmental and did not rely in a single approach for all staff (Melton et al., 2010). Flightgate had been used in a published change programme with Mental

Health and Learning Disability services across a health board in England and was based on well articulated research describing mechanisms or catalysts for change, which included a strong focus on ‘context’, which had congruence with my own philosophical stance outlined previously. Within our wider network, the team had the opportunity of free access to and training in the Flightgate model.

The pragmatist perspective, permits the researcher freedom to choose methods, techniques and procedures which best meet their needs. At the time, I took a ‘pragmatic’ decision, together with the research team to adopt Flightgate, because it was not only available to us but I had confidence that this approach had a strong chance of success in supporting a large number of practitioners across a range of different teams to identify their own and their service needs for support to change in adult autism diagnostic services.

Although we were interested in applying this framework to reduce wait times, this study did not set out to explicitly measure which aspects of the process, practitioner factors or mechanisms influenced outcomes. The study draws on implementation science, through application of practice development theory rather than this being the focus of the research. Future study to measure the organisational and individual practitioner factors and how these influence wait times could support more specific tools for implementing improvements to autism assessment and diagnosis.

Leadership to support service change

The success of the work implementing changes and reducing wait times within adult services (paper 4), motivated me to use the research skills acquired in this process and to undertake further related research through applying for a small grant to lead a local collaboration. As a practitioner and researcher, embracing the challenges studied in the research to understand delays in diagnosis, I sought permission to lead a programme of change within my own clinical service (for children), which was also focussed on addressing waiting times. This programme, delivered through multi-disciplinary collaboration, was effective in improving waiting times for assessment in child services over a 2 year period of data collection (paper 5).

In extending the approach trialled in paper 4 into my own work setting, with a much larger group of staff (paper 5), I was also interested in giving consideration to the importance of leadership roles to support the dissemination of innovation. Whilst some staff within this health board were motivated by the opportunity to focus on reducing autism assessment wait times, others had different priorities and were less motivated to engage. Within the NHS leadership academy (2013) framework, two elements described are key talents for leadership

in health supporting meaningful translational research (Shickle et al., 2014), those of “connecting our service” and “influencing for results”. Within my own role in the NHS team, I sought to connect to and create shared agendas with stakeholders, to understand unspoken needs and agendas and to act flexibly but also strategically to overcome obstacles. My role was to provide rigorous, thorough and robust evidence, synthesis of information and reasoning as I led the programme of work. As well as the establishment of a strategic pathway planning group, with representation across the geographical spread of the area and across professional groups, the leadership role required persistence and re-iteration of key messages and agreed actions to colleagues, from different professional backgrounds and cultures, over time to support their practice change. Once again, the intersubjectivity and iterative nature of this mixed method approach appealed to my ‘inner pragmatist’.

In this highly complex healthcare intervention (paper 5), focussed on service improvement (Pawson et al., 2014), a dynamic range of solutions were applied. The intervention programme included strategic and organisational change, for example, the three main diagnosing professions (CAMHS, Community Child Health and AHPs) were asked to operate under a single ‘autism’ pathway for children aged 0-18, triaged collaboratively, for the first time. There was a significant role change for staff; the new pathway meant that 25% of diagnostic assessments would now be carried out by local practitioners, with a move away from the ‘expert’ model. Staff who did not see themselves as ASD experts, would now have a greater role in diagnostic assessment. A triage process was implemented to channel the more complex cases to expert practitioners. This required expert practitioners to take on a greater leadership role including providing training, mentoring and support to colleagues. There were procedural and administrative changes, for example, staff were asked to stop doing school observation visits for all children and only to do this where was discrepancy between home and school standardised assessment scores, based on evidence gathered about effective and efficient use of time. Data collection and appointment management was adapted to suit the new pathway and staff needed support to take on new ways of working. The outcomes and progress with each of these aspects of the programme were reviewed through a multi-disciplinary pathway management group as an additional leadership element of the programme. In this work, following reflection on gaps in the work in paper 4, end-user feedback was sought and shared with local service providers and managers to support understanding of the variables which had mediated the wait time reduction reported in paper 5 (King et al., 2018), which summarised recommendations for ongoing service development based on this feedback.

In conclusion to research programme 2, although it is well known that making real world impact with research is challenging. Papers 4 and 5 report success in bridging the research – practice gap, which resulted from and was embedded into the methodology underpinning the study. The outcome was to be one of the first research programmes to evidence reducing the delays in ASD diagnostic assessment in child and adult services and completes the body of work about waiting times, whilst setting the stage for further development of my research.

Improving outcomes through evidence based interventions

There is an increasingly strong narrative amongst autistic people and the broader autism community in support of expecting neurodiversity (Singer, 2017) and against the notion of trying to ‘cure’ autism (Gillespie-Lynch et al., 2017). I am pleased that this position strongly resonates with the pragmatist perspective and position of speech and language therapists and others who have approached developmental differences from a social pragmatic perspective (see below). As is the case with the issue of diagnosis, the pragmatist stance is that intervention occurs in the ‘doing’ of actions, which have contextual relevance, and meaning for those receiving or taking part in interventions. I and many others advocate universal inclusive approaches, targeted approaches and reasonable adjustments within everyday naturally occurring social and physical environments to enable the successful and meaningful participation of autistic people in daily life (World Health Organisation, 2007).

The position that support and intervention should be based on need and not diagnosis is therefore absolutely valid. It is equally valid to accept that relevant ASD specific interventions and supports are more likely to be accessible following diagnosis (Begeer et al., 2013) and families are in a better position to focus their time and access supports when there is diagnostic clarity. Clinical guidelines (NICE, 2011; 2012; SIGN, 2007; 2016) recommend a range of interventions and a strong body of literature supports the notion that timely diagnosis is crucial for access to relevant interventions. However, unless individuals can access useful and effective supports and interventions to improve outcomes, diagnosis of ASD may be of limited benefit.

In my research role, and following my participation in the SIGN guideline writing group, I was interested in engaging with research to support the translation of clinical guidelines into practice for specific interventions, recommended and commonly delivered amongst my own community of practice. On completion of programmes 1 and 2, research programme 3 (paper 6) permitted a shift of focus, to the important issues surrounding the provision of supports and interventions which contribute to positive outcomes in autism. There are many assumptions,

explored below, surrounding the questions of how this work fits within a pragmatist framework, what outcomes are important, which interventions are most effective and relevant for individuals and their families across the lifespan and how practitioners and services should prioritise resources.

I have shared my pragmatist stance and the ways it underpins my research work, however pragmatism has also provided a strong basis for my clinical perspective and interests in social pragmatic approaches to intervention, including my decision to select parent focussed interventions as an area of focus in paper 6.

A social pragmatic approach

ASD is fundamentally a social impairment (Mandy et al., 2018), in which individuals are known to have a great deal of heterogeneity (Masi et al., 2017), including a common difficulty with generalisation of skills from one context to another. There is an acceptance that the social environment (knowledge, actions and attitudes of people around the child) has a strong influence on participation in children with a range of additional support needs including autism (Maciver et al., 2019).

There is a range of views and a longstanding, and at times heated, debate with those advocating discrete trial training and applied behaviour analysis on one side and advocates of the ‘social pragmatic’ position advocating interventions in naturally occurring environments (Prizant and Wetherby, 1998; Binns and Cardy, 2019) on the other. The SIGN guideline found evidence for approaches across this debate; however, it is important to continue to question the measures used to report positive outcomes. Just because we can measure something does not mean that we should. With some research, it is clear that if you teach a specific skill you can prove that it improved (Dillenburger and Keenan 2009). From a pragmatist perspective, this does not tell us whether this was the important thing to target for an individual in context now and in the longer term. This is an ongoing debate and in developing my own position, I am conscious of my desire to contribute to the development of outcome measures that measure what we should measure as important in outcomes for autistic individuals and their families (McConachie et al., 2015).

From my position and that of the social-pragmatists, it is essential that autistic people are supported with social communication in real world contexts with the people they are most motivated to interact with and who are most frequently available to them. This is more often than not, their family and for many, parent mediated interventions or parent focussed interventions provide this opportunity. Families of children and adults with autism continue to

express dissatisfaction with the level of support received (e.g. Crane et al., 2015) and in the current political climate, public services have resource limitations and are under pressure to use resources effectively. Good quality evidence about which interventions work for whom could support families to experience greater support and support services to use resources available effectively.

Given the additional strong evidence of high levels of parent stress and negative impact on quality of life for parents of autistic children (as outlined in paper 6), there is considerable evidence of the need for research, to support practitioners with evidence to inform them about what parent interventions are available, which interventions work, through which mechanisms and in what contexts.

The evidence available for review is relatively strong for the pre-school stage (SIGN 2016) but becomes increasingly weak over the age span and indeed, it is likely that key social communication opportunities as children get older may equally occur within education contexts, raising further questions about how the evidence for the pre-school stage is applicable for older children and adults.

Paper 6 draws together several inter-related elements, which in themselves require discussion, including:

- What are parent focussed interventions?
- Why focus on older children and adults?
- What methods are appropriate to answer the research questions
- What outcomes are relevant and how these are measured
- How should we interpret effect size in the context of the studies available

What are parent focussed interventions?

Parent focussed interventions are amongst the recommended psychoeducational interventions for ASD (SIGN, 2016; NICE, 2011; NICE, 2012) which impact reciprocally on the wellbeing of the individual with autism and their family. They involve professionals working with parents and or carers to enable them to make adaptations, which are intended to be supportive of their autistic child. There are a range of intervention types included under this heading, described further below. Clinical guidelines do not provide clarity about which specific interventions or elements of interventions are most effective for different individuals and

across the lifespan. The range of interventions included in the systematic review is outlined in paper 6.

Why focus on older children and adults?

Within this thesis I will explore in more depth, the reasons for selecting the age of 7 years as a cut-off point and benefits and challenges of grouping different interventions under the umbrella of ‘parent-focussed interventions’.

Pre-school children

Services regularly deliver evidence based support for pre-school children with ASD through parent mediated interventions. Improvements in child and parent outcomes are reported in a Cochrane review and several RCTs, which refer to children up to the age of 6 years (Oono et al., 2013; SIGN, 2016). Oono et al. (2013) identified a small but significant effect size in improvements to parent-child interaction following intervention. A recent meta-analysis included 19 RCTs on this topic (Nevill et al., 2018) also identified positive effects of such interventions, however outcomes were focussed solely on child skill variables. In the field of ASD, very few interventions have this level of consistency in intervention delivery across studies or evidence supporting their effectiveness, Given the strength of this evidence, relative to evidence for similar interventions for older children and adults, I elected not to review intervention research at the pre-school age range (up to age 6 years).

Older children and adults

Parent or family focussed interventions are similarly recommended in clinical guidelines for school aged children and adults; however, this recommendation is currently based on fewer studies and a much lower level of evidence. There have been two recent systematic reviews related to this topic in children (Kuhaneck et al., 2015; Cuchia et al., 2016) but no meta analysis was reported, with no reviews of interventions for families of adults, despite the increased likelihood that parents of individuals with ASD will still live with their child in adulthood (Karst & Van Hecke, 2012).

No systematic reviews looked specifically at the effects of parent focussed (ASD) interventions for older children or adults, supporting my decision to follow this discernible focus for paper 6.

Intervention types

While parent mediated interventions for pre-school children were well defined, parent focussed interventions for older children and adults were not. For this reason, I was motivated to undertake research to build what I saw as important evidence to support the further refinement of our understanding of what constitutes a parent focussed intervention for older children and adults, providing recommendations for practice and future research.

There may still be professional debate about whether it is valid to group the different interventions included here and it is important to justify my decision to do so. Although there are differences between interventions, which may in future lead to them being conceptualised as different interventions, they do have much in common. They are delivered through expert practitioners teaching parents additional skills and knowledge, to better understand their child, to do more of what works well or to make helpful changes to daily routines (e.g. Kaminski et al., 2008; Kuhaneck et al 2015; Cachia et al., 2016). They all aim to improve parent wellbeing through a short term focused opportunity. Parents derive benefits of meeting others in a similar position.

I made the decision to group interventions sharing these characteristics, in order to permit meaningful consideration of the evidence. Four parent focused intervention sub-types were identified prior to the study (which share the above commonalities) and are described further in paper 6.

1. Parent education, training or coaching programmes
2. Mindfulness or relaxation training
3. Parent support groups
4. Multi-component child and parent intervention models

As a pragmatist, I do not see this as the end of the discussion. I hope that by first setting a line in the sand, reviewing existing research and sharing my own definition, based on the current evidence, that practitioners and researchers in the field can continue to refine and reach consensus on ‘parent focussed interventions’ and how we should measure their effectiveness.

A clinical perspective

Within my clinical role, I was aware of the significant proportion of our service resource given to interventions and the positive responses from families to supports and interventions

accessed through local services. I was particularly driven by my experience of delivering parent mediated interventions for pre-school children. Although parent supports for older children were also being provided, I was aware that there was less evidence for these. There was a clinical need for research evidence to underpin practice in parent focussed interventions for school aged children and adults.

Through my active involvement in the SIGN guidelines (SIGN, 2007; 2016), I was acquainted with the current evidence, and the extent of the knowledge gap. Although SIGN have well defined standards for reviewing the quality of research which can be included in a clinical guideline (SIGN, 2015), the lack of evidence around ASD interventions meant that some guideline recommendations were based on expert clinical opinion rather than on strong evidence, including the recommendation for parent focussed interventions to be offered across the lifespan.

Research methods for evaluating intervention research

From an epistemological perspective, one could argue that SIGN and the system of grading evidence in clinical guidelines prioritises quantitative research over qualitative, with the highest levels of evidence considered to be meta-analyses and randomised controlled trials (RCTs) followed by systematic reviews and then cohort studies. However, for a clinical researcher interested in a mixed methods approach, this is not necessarily problematic. The work already outlined, on diagnostic waiting times began with quantitative data gathering, which required the definition of concepts to be measured and the development of shared language, truths and understanding of key variables. For that study, there were no relevant meta-analyses or RCTs to answer the research questions and too few papers to make systematic review possible. The guidance on wait times in clinical guidelines was largely based on expert opinion and this therefore determined the research methods applied. As a pragmatist, research methods selected to answer a question should take account of such context.

In the case of developing methods to answer questions about the effectiveness of ‘parent focussed interventions’ the context was different and therefore I had the opportunity to apply different research skills in the form of systematic review and meta-analysis.

Systematic review

Systematic reviews have an important place in healthcare and are pivotal to recommendations made in clinical guidelines, both synthesising evidence across research studies and

highlighting gaps in evidence (Shamseer et al., 2015). They begin with a focussed question and follow internationally developed PRISMA guidelines (Moher et al., 2009), which provide guidance on a systematic approach to identification, selection, appraisal and synthesis of evidence in research studies. For this reason, systematic review seemed to be an important research skill to apply as well as the most appropriate method of answering the question posed in research programme 3 (paper 6).

Meta analysis

Studies incorporating the statistical technique of meta analysis are equally pivotal in clinical guideline development and are viewed as the highest level of evidence. Within interventions for ASD, these are relatively rare (NICE, 2011). Meta analysis, preceded by systematic review, combines data from multiple studies, in order to generalise results from similar experiments, across a larger sample size than single studies alone (Clark-Carter, 2019). For example, those that test ASD parent interventions. This is done, in order to increase the potential to determine whether a real intervention effect exists and whether it is statistically significant, i.e. to increase power to detect small effects that a single study is not able to determine (Higgins & Green, 2011).

As described in the Cochrane Handbook (Higgins & Green, 2011), one of the fundamental principles of a meta analysis is a two-step process that involves the computation of a summary statistic for each study to demonstrate the observed treatment effect. This is followed by the calculation of a weighted average of the summary statistics estimated in the individual studies to obtain an estimate of the pooled treatment effect across all studies. Randomised controlled trials (RCTs) characterise the gold standard for the development of evidence-based medicine (Castillo et. al., 2012), since these are immune to internal bias by design (Borenstein et. al., 2009). The Cochrane Handbook (Higgins & Green, 2011) states that potential biases tend to be relatively greater in non randomised studies (NRS) than randomised studies, especially as a result of selection bias and reporting bias.

However, according to the Cochrane Non-Randomised Studies Methods Group in the Cochrane Handbook (Higgins & Green, 2011), including non randomised studies in a review may be justified if an adequate number of randomised trials cannot be determined. Nevertheless, it is firmly advised to not merge evidence from RCTs and NRS. Instead, a particular review might constitute ‘component’ reviews that involve different study designs, and if both RCTs and NRS are incorporated, these should be demonstrated independently, as I have done in paper 6.

Meta analysis is conducted separately for RCTs and NRS. NRS included case-control trials (consisting of two independent groups i.e. experimental and control) and single group pre-post trials (consisting of one group and its scores pre-intervention and post-intervention). Meta analysis of single group pre-post trials could not be performed in paper 6, due to lack of reported data (S_{diff} and r were not reported). Taggart (2001) asserts that if the included NRS are fairly resistant to biases and considerably homogenous in that respect, meta analysis may be used to synthesize data across these studies. Furthermore, the Cochrane Handbook (Higgins & Green, 2011) also confirms that effect measures used in meta analysis of RCTs can also be used in meta analysis of NRS. Although larger number of studies permit greater meaning being given to results, it is possible to conduct meta analysis with 2 studies or more (Borenstein et al., 2009).

The meta analysis procedure undertaken starts with the computation of effect size across studies; this is the standardised mean difference (Hedges' g). The next stage in analysis involved fitting a fixed-effect and a random-effects model along with the corresponding forest plots. Although both models were initially implemented in the analysis, the random-effects model should be favoured whenever possible. This is because studies used in this analysis are related to an extent that it may be sensible to combine knowledge from them, but they cannot be assumed to be identical in a way that they would share the exact same common true effect (as assumed by a fixed-effect model).

The selected studies were conducted by clinicians separately and hence cannot be deemed to be functionally identical. There are contrasts in a range of variables, such as participant characteristics or intervention usage, resulting in different effect sizes underlying different studies. The random-effects model takes into consideration both within-study and between-study variation and is therefore more justified in this scenario. Moreover, as mentioned by Borenstein et al., (2009), results from a random-effects model are generalisable since it does not assume the exact same, narrowly defined population for all studies. This makes it possible to extrapolate results from the identified population to a range of scenarios.

I took some time over the decision to undertake meta analysis or not with the small number of studies, with adequate data to permit their inclusion. However, I concluded that the meta analysis planned did meet guidance reviewed above; and given that clinical guidelines already recommend parent focussed interventions across the lifespan, that this evidence would still be relevant and support future research in this field.

Parent focussed interventions were already recommended (SIGN, 2016) based on several pre-existing RCTs and a Cochrane review relating to parent mediated interventions for pre-school children. The evidence for such interventions for older children and adults was a clear gap. Based on my own scoping review, a systematic review and meta-analysis would be a good place to start in answering the research questions, however, there would still be the need to explore assumptions and definitions related to inclusion criteria and parent wellbeing focussed outcome measures, rather than child skill focussed measures. The complexity in ASD interventions and in related research has contributed to an inadequate (although large) body of research from which to draw conclusions about the effectiveness of interventions (Warren, 2011). The variables affecting the manner in which ASD focussed interventions result in generalisation of new skills across contexts and have impact in the longer term is difficult to measure for a range of reasons (McConachie, et al., 2015).

Outcomes for ASD specific interventions – what should we be measuring and how?

The further element for consideration in this critical appraisal, is ‘outcomes’ and the consideration of what should we be measuring and how? There is a gradual shift in the focus of outcome studies, towards slowly but increasingly including measures of wellbeing and participation in meaningful daily life experiences. However, this shift in perspective is still in motion and the research in paper 6 adds valuable evidence to support the need for change in outcome measurement in ASD.

Although McConachie (2016) identified that families rated constructs of emotional wellbeing and effects on the whole family as the most important outcomes to be measured, professionals reported that they measured the things they had tools for (e.g. skills in play, motor development, speech, social interaction) rather than outcomes for wellbeing for parents. Despite the availability of tools to measure wellbeing, this outcome is still reported with limited frequency in intervention studies (McConachie, et al., 2015). The field is at a stage where there is recognition that we should be measuring wellbeing outcomes but there is a lack of clarity over how this should be done.

Wellbeing of families of people with ASD

In systematic review, the researcher is reliant on previously published studies and for this reason I was reliant on ways in which others have conceptualised and measured wellbeing in intervention studies. Wellbeing is characterised by two key facets of being subjectively happy or feeling good and functioning well in relationships with others and autonomy, self-acceptance and feeling competent (Stewart-Brown & Janmohamed, 2008; Wahlbeck, 2015).

Reported wellbeing outcomes from parent focussed interventions reviewed include improvements in: parent wellbeing (Ekas et al., 2010; Oono et al., 2013; Suppo & Floyd, 2012); communication and shared parent-professional understanding; generalisation of learning and behavioural and academic outcomes for children (Matson et al., 2009; Benson, 2015). To provide focus in a complex field, I opted to focus on the first of these.

During the review (paper 6), I identified five commonly reported parent wellbeing measures: quality of life, parenting stress, parent self-efficacy, parenting style and parent satisfaction. This is not an exhaustive list, but given that variety and inconsistency have thus far made it hard to compare data across studies (Kuhaneck et al., 2015), I opted to focus on these five as the most commonly reported measures, which have relevance to autism parent focussed interventions.

Quality of life

Quality of Life (QoL), based on Schalock's (2004) widely adopted framework, is defined with reference to dynamic and complex factors from a range of domains used to assess individual perspectives on participation in daily life through the eye of the experiencer. These are a) macro-societal: how the external environment and socio-political makeup of society provides community based, social support and resources and b) micro-individual: including physical and mental health and wellbeing, psychological outlook, role in society, independence, autonomy and perceived control over life, material and financial circumstances. (Brown et al., 2004, p46).

There is consistently reported poor QoL in families of people with ASD (Boehm et al., 2015; Eapen & Guan, 2016; Vasilopoulou & Nisbet, 2016). QoL in families of individuals with ASD is not only lower than for families of typically developing offspring but also lower than families with a member with other disabilities (Eapen, 2016). There is therefore strong evidence that family QoL and family systems are affected by having a child with ASD (Eapen & Guan, 2016) and some evidence that well supported families are in a better position to support their children (Russa et al., 2015). We can infer that family QoL for parents; carers or spouses of adults with ASD are similarly affected. Notably the reported systematic reviews have not considered QoL in relation to intervention outcomes.

Parenting stress

QoL and parenting stress are closely related constructs. Schalock's (2014) work on QoL and Perry's (2004) model of stress in parents of children with disabilities have advanced our

understanding of a social model of evaluation of outcomes for individuals with disabilities (Verdugo et al., 2005). Over 90% of parents of children with ASD experience substantial parenting stress (Nikmat et al., 2008), which is higher than for parents of typically developing children or children with other disabilities (Watson et al., 2013; Bendixen et al., 2011; Cachia et al., 2016). The earlier interventions starts, the greater the reduction in parent stress (McConachie & Diggle, 2007).

Parent self-efficacy

Parent self-efficacy is another QoL related concept (Ji et al., 2014) affecting the quality of caregiving (Benn et al., 2012), caregiver sense of competence (Ji et al., 2014) and parental confidence (Whittingham et al., 2009a) commonly measured in the Parenting Sense of Competence Scale (Gibaud-Wallston & Wandersman, 1978). An interrelationship exists between parent self-efficacy and child education success, behaviour and wellbeing (Sanders et al., 2005; Sofronoff et al., 2004; Whittingham et al., 2009b; Benn et al., 2012; Ji et al., 2014). This is a particularly relevant measure for interventions aiming to ‘upskill’ parents of autistic children and to increase their confidence in parenting their ‘child’ who is not following a typical developmental trajectory.

Parenting Style

Parenting style is another multi-dimensional construct, which encompasses a number of different constructs, which have a dynamic relationship with child behaviour (Neel et al., 2018), measured in studies selected, using the ‘Parenting Scale’ (Arnold et al., 1993), with measures of ‘laxness, over-reactivity and verbosity’ (Whittingham et al., 2009a).

Parent satisfaction

Parent satisfaction with interventions was also measured in the studies reviewed. Reviewing the experience of participants in an intervention is good reflective practice (Brown et al., 2018). Ultimately, if parents are not satisfied with the intervention they are less likely to engage and it is less likely to be effective. As all the interventions reviewed are at a relatively early stage of development, this is an important measure. However, it is equally important to consider that just because parents enjoyed being part of an intervention or liked the people they met – this does not necessarily make it effective in providing the most relevant and useful strategies for parents of a child with ASD. Studies used a range of means to measure parent satisfaction

Quality assessment

Within my own systematic review (**paper 6**), it would be important to assess methodological quality of selected studies to objectively account for the quality of evidence reviewed including risk of bias. The Cochrane Collaboration provide a range of resources for this purpose, including their tool for quality review of RCTs (Higgins et al., 2011). In researching tools for quality assessment of observational studies, I identified the Effective Public Health Practice Project (EPHPP) Quality assessment tool for observational studies (Thomas et al., 2004).

Selecting studies for review

Within the systematic review, selection criteria were identified, before the planned search strategy was undertaken, to comprehensively search electronic databases, as outlined in paper 6. Studies met the following inclusion criteria, devised using the PICOS approach (Liberati et al., 2009):

1. An intervention study focussed on parents/carers of children (7-18) or adults (over 18) with Autism Spectrum Disorder
2. the study reports parent outcomes using standardised assessments of parent wellbeing
3. the quantitative research design includes a non-intervention or pre-post comparison group
4. the paper was published in a peer reviewed journal, in English. Qualitative studies, review papers, non intervention studies and intervention studies without parent participation were excluded, as were parent focussed interventions for individuals without ASD or for younger children.
5. Where participants were across the age range, studies where the mean age was below seven were excluded and studies with the mean age above 7 were included. Grey literature was excluded, for example, technical reports, dissertations or unpublished documents.

A final word on epistemology

In critical appraisal of underlying assumptions in paper 6, I have discussed how my pragmatist position influenced the subject of enquiry and I will also consider my own epistemological position in relation to research methods applied. Systematic review and meta-analysis are used

to build new evidence from connections between and across different published studies. Although, these could be seen as quantitative approaches derived from and sitting within a positivist tradition, in actual fact I would argue that they are also core tools for a pragmatist researcher, defining knowledge through its meaning in context to those using it. Suri (2013) clearly articulates alternative epistemologies, which are equally valid for systematic review and meta-analysis.

The positivist approach has a focus on objective, unbiased, universal truths which are decontextualized and in this field run the risk of a false sense of accuracy and certainty. The world of ‘parent focussed interventions’ as highlighted here, does not currently include clear objective definitions of concepts being measured but rather, it is a field where I seek to make connections between related research studies to develop a deeper and more nuanced understanding of the subject. As a pragmatist, I am interested in the data available, as it stands but also wish to continue to understand contextual variables and be open to an iterative process and future reconfiguration of key constructs in relation to how such interventions and wellbeing outcomes are defined.

In using meta-analysis, I have asked myself whether I am colluding with the predominant narrative that quantitative data and the positivist position is somehow better than other research methodologies. However, I have continued to take a critical approach in how the meaning of data and specifically effect sizes, are interpreted (Schäfer & Schwarz (2019). Effectiveness in intervention studies is commonly measured empirically through reporting of effect sizes, to allow comparison of different interventions, using different sample sizes. However, in meta-analysis, the value of this is not black and white and is relative to the quality of the studies being compared. In my own publication, I was keen to highlight the limitations in the findings, of medium effect sizes but with a small sample size and relatively low quality studies. The low effect size reported in studies of parent mediated interventions for pre-school children may in fact be interpreted with greater confidence. This is because of the higher quality of studies included when compared to the medium effect sizes reported in paper 6, with fewer high quality papers and small sample sizes in studies of parent focussed interventions for older children and adults. This observation will be considered in terms of the discussion and conclusions from my research at this point.

In research programme 3, I have begun to engage with a range of complex concepts and research methods, which are important to the development of a common framework for evaluating autism focussed parent intervention outcomes and hope to continue to be an active researcher in this field as it evolves.

In summary, the methods section of this critical appraisal has included consideration of epistemology, reasoning and decision making related to mixed methods applied across six different publications and three separate research programmes outlined on page 7-8. I hope that it provides evidence of breadth and depth in research and critical thinking skills at doctoral level. The methods section has included reference to international literature and consideration of concepts and theories underpinning the work, as well as highlighting areas where there are gaps in our current knowledge. Results are shared below, where I can report on research data together with the impact this has had on practice across Scotland. Through publication and wider dissemination of results there is also beginning to be international recognition of the relevance of this work to countries outside of Scotland. The approach taken has led to exciting results, which perhaps against the odds, bridge the research-practice gap as a result of the aims and methods applied. As with all research there are strengths and limitations and there is the potential to continue to build on this work in research, policy and practice.

Results

The wide ranging results from this work are detailed in the 6 published research papers and for the purpose of the thesis, are presented and discussed in summarised form below.

Waiting for diagnosis

Papers 1-3 reported the current wait times for diagnosis and provided quantitative and qualitative data to explain the reasons for these waits and possible solutions. In Scotland, although adherence to evidence based clinical guidelines was found to be high (McKenzie et al., 2016a, 2016b) and was unrelated to waiting times (paper 2), the research found that there is currently a long wait for diagnosis of ASD, with 74% of child and 59% of adult services exceeding the 119 day standard (McKenzie et al., 2015). The reported mean age of diagnosis in children has not reduced over the last decade (Brett et al., 2016) and data reported is often dependent on the age group or learning level of the cohort studied, with the mean age of diagnosis ranging from 38-120 months.

No previous study was found which specifically set out to report age of ASD diagnosis in adults. However, it can be inferred that any diagnosis of ASD in adulthood represents delayed diagnosis of this developmental disorder of childhood and recent studies report similar mean ages of diagnosis regardless of gender, ranging from 31 years to 34.1 years (Rutherford et al., 2016a). The research presented here found that the mean age of diagnosis in Scotland was very

similar to national and international reports, at 8.9 years (107 months) for children (McKenzie et al., 2015) and 31.6 years for adults (Rutherford et al., 2016a). Interestingly paper 1, focusing on gender, found that the delay for females occurs prior to referral and not during the assessment process, which has key implications for allocation of training and resources to address this challenge. Paper 1 has been to date the most cited of this collection of publications, despite being a brief paper. This highlights the importance of the pragmatist approach, which was cognisant of issues pertinent to the community of practice, evidenced through publishing data and findings felt to be timely and clinically relevant, even though the data did not involve complex analysis.

Throughout this thesis, I have highlighted the decisions made in planning and delivering the research, from a pragmatist position, which are intended to support the delivery of research relevant to practice and thereby supporting the uptake of the research in practice, through dissemination strategies. The publications are evidence of dissemination to an international audience and encouragingly our research has been referenced in the SIGN (2016) and Australian Autism Clinical Guidelines (2018). This provides evidence of the relatively quick pathway to impact, which may have arisen because it is work rooted in a key question of interest to guideline writers, where there is a dearth of evidence.

Reducing the wait through evidence based practice change

The research involved direct dissemination of findings nationally through the work published in papers 4-5 and we were able to demonstrate that waits in both adult and child services could be significantly reduced, to within recommended time standards by applying the solutions generated in papers 1-3, together with a locally focussed change programme. For example, the programme in paper 5 led to a statistically significant reduction in wait times from referral to diagnosis from 270 days to 122.5 days ($t(20) = 5.5, p < 0.05$). As well as reduced waits, there was an increase in the proportion of girls identified, suggesting that the strategy employed for raising recognition of girls might be effective.

On reflection, there were strengths and limitations in the decision to use Flightgate as a practice development framework (paper 4) but not to explicitly undertake measurements of implementation processes and mechanisms. Since the completion and publication of papers 4 and 5, the field of implementation science has continued to expand and I am interested in using this opportunity to further reflect and consider future possibilities opening up to clinical researchers interested in change frameworks.

As a result of this research (papers 1-5) I was able to further elucidate key components of effective ASD service improvement, with a focussed and supported action plan based on literature review and the research experience. These were:

- **The importance of good data** to keep staff informed about wait times and to support service planning in relation to demand, capacity and efficiency of the service provided. Prior to the programme of change there was no consistent way to know how many referrals were made for ASD assessment or how long they waited. Estimates made by service leads were later found to be inaccurate. They underestimated the number of referrals by more than half.
- Having and following a written, locally relevant, **evidence based ASD Pathway**, such as the one developed and reported in this research.
- **Flexible approaches to assessment**, where ‘one size does not fit all’, so that complex cases are managed differently to more straightforward cases, thereby making more effective use of resources. Although in our research, services were not found to take account of individual case complexity in their autism assessment pathways, I have been able to explore this concept further here. I hope that it becomes part of our ongoing dialogue in practice because of its important in supporting the development of more effective pathways and applied in the work reported in paper 4 and 5.
- **Implementing referral management and triage**, to reduce duplication and actively manage the range of referrals received, according to the pathway guidance.
- **Reflection on service configuration** – with triage teams to consider case complexity alongside the optimal skill mix of the team allocated the case, to reduce duplication and wait times.
- **Training and Mentoring of Staff** implementing the new ASD pathway, based on the NES (2014) Autism Training Framework, with a focus on a different approach for each of the four levels of autism knowledge - informed, skilled, specialist and expert, as outlined in paper 5.

It is anticipated that reporting these results will support practitioners around the world, seeking to address the challenge of demand and capacity for diagnostic assessment.

Systematic review of parent focussed interventions

The final set of results reported relate to systematic review and meta-analysis of intervention studies about parent focussed interventions for older children and adults. This research began by identifying and proposing definitions for key concepts, such as what is meant by the term 'parent-focussed', what interventions and outcomes are reported and how are these measured. Following systematic review and meta-analysis, I identified that parent focussed interventions are heterogeneous interventions with no single intervention being reported more than once.

Of the 57 full text articles reviewed, 22 studies were identified, of which 5 were RCTs, 3 non randomised controlled trials and 14 cohort studies, which met inclusion criteria for the study. There were 22 different interventions named and 20 different wellbeing measured reported. In the quality assessment, undertaken using the EPHP tool (Thomas et al., 2004) two studies were given a strong rating, 15 moderate and 5 were weak.

Through the systematic review in paper 6, it was possible to clarify that parent interventions are recommended for a number of reasons: firstly, because a diagnosis based on difficulty with social communication is inevitably context dependent and parents are present more often within daily contexts than professionals. Secondly, parents are very well placed to provide the frequent opportunities needed to support learning and progress with their child. Thirdly, difficulties with generalisation are inherent in ASD and this is always a risk with interventions which teach a child something in one context and expect them to use it in another. For changes to be meaningful, learning that takes place in the context it is needed, is most effective. Fourthly, parent stress and parenting style have a reciprocal relationship with the behaviour of the child or adult with ASD and therefore these are important areas to target in intervention, especially given the high percentage of adults with ASD who continue to live with their parents. Finally, compared to many interventions, parent focused interventions are likely to be cost effective, with relative ease of delivery, although further research is needed to evidence this. In the recent Scottish 'microsegmentation' report, which has analysed the financial implications of autism, one of the recommendations is that more focus should be given to parent supports and interventions, as a way of reducing societal costs of ASD (Mackay et al., 2018).

Meta-analysis results

Outcome measures were heterogeneous in paper 6. Although the sample size was small and should be interpreted with caution, separate meta analyses compared five wellbeing outcomes (quality of life, parent stress, self-efficacy, style and satisfaction), within each of two

intervention types (mindfulness and parent education and training), where data permitted inclusion. Data for parent support groups and relaxation could not be included in meta analysis, as information to permit calculation of effect size was not reported. Throughout paper 6, I have made it clear that results should be interpreted in the context of the small number of studies.

Through meta analysis in paper 6, three statistically significant outcomes were obtained. These support the assertion that parent focussed interventions in ASD can be effective in improving parent wellbeing.

1. For reducing parent stress via mindfulness training ($g=-0.52$ [-0.98,-0.07] with a medium effect size ($n= 2$ studies))
2. For improving parenting style through parent education (laxness, $g=-1.00$ [-1.5,-0.49], verbosity $g=-0.97$ [-1.34,-0.60], over-reactivity $g=-1.00$ [-1.38, -0.61]) ($n= 2$ studies)
For the three elements of parenting style a large negative effect size was found, suggesting the intervention reduces these behaviours in parents.
3. For improving parent satisfaction through parent education ($g=0.70$ [0.40, 1.01]) there was also a large effect size, suggesting the interventions improved parent satisfaction ($n= 5$ studies).

This outcome, with evidence suggesting that parent focussed interventions can be effective, adds weight to the recommendations in ASD clinical guidelines over the use of some types of parent interventions in ASD, which have the potential to reduce parent stress, and improve parenting style and satisfaction. These are parent education training and mindfulness.

Interpreting effect size

Although writers of clinical guidelines should be able to interpret and interrogate published data, clinicians may not have confidence or guidance to interpret effect sizes (Ferguson, 2016) and therefore the narrative surrounding the reporting of results is important. Although to the naïve observer, a large effect size may suggest that an intervention is more effective, useful or important than one producing a small or medium effect size, in actual fact Simpson (2019) and others would argue strongly that this is not that case as the effect size does not measure relative effectiveness.

For this reason, results and effect sizes must be interpreted with an understanding that they are ‘estimates’ of true real world effects and that bias can be introduced through small sample size and non randomised sampling. It is important to highlight that, in the quality review of the

studies, only two were identified as of ‘strong’ quality, together with the context that no single intervention was included in more than one study. By way of comparison, although meta-analysis of parent mediated interventions for children under 6 years (Oono et al.2013; SIGN, 2016) reported small (and significant) effect sizes, they included a much larger sample size and greater consistency in the interventions included. For other parent interventions, where no significant effect was reported, there may be a need to re-assess the quality of research before ruling them out. It may be that they are effective but that this is not captured by the design of the research applied.

Effect size alone should not be used as the means of recommending an intervention. As a pragmatist I would like to further develop a more nuanced approach to understanding and interpreting the research currently available, as well as making a contribution to future research which is of ‘stronger’ quality and addresses some of the current limitations in the field.

Recommendations for future research

Increased value in future research into parent focussed interventions can in part be addressed through research methods, such as:

- using larger sample size,
- randomised designs,
- better definition of interventions,
- consistency in outcome measures,
- inclusion of ASD specific and standardised wellbeing measures,
- use of replication studies, and
- use of mixed methods studies which give the participant perspective to interpret quantitative data.

I have reached the conclusion that there is a need to develop new, theory driven, manualised interventions tailored to the needs of parents of older children and adults. However, a key missing element is a shared theoretical framework for ASD parent focussed interventions underpinning the research to provide contextual relevance and support comparison across studies. Such a framework could support our understanding of which elements of the intervention are associated with particular outcomes, for particular individuals.

A theoretical framework

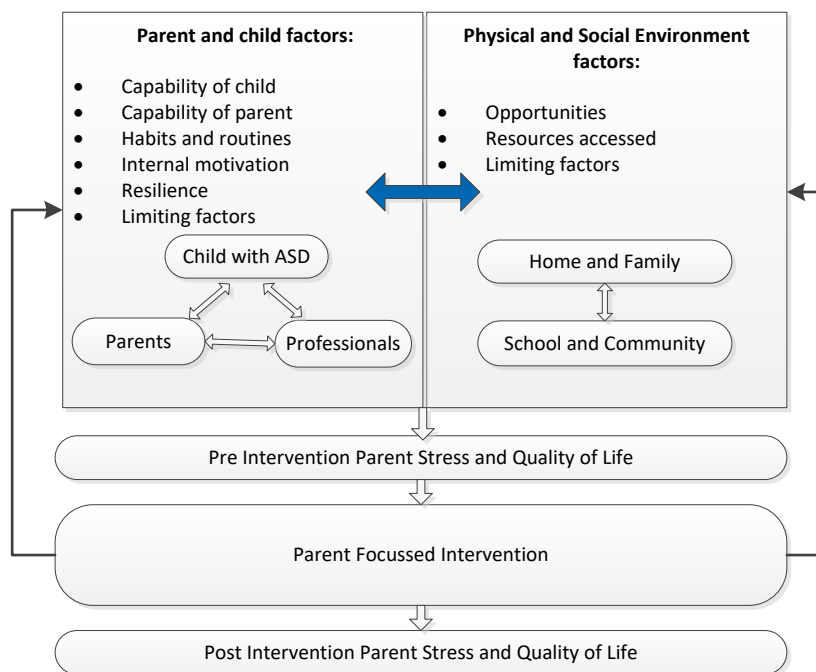
Autism research is an exciting and emerging field with the potential for positive impact on daily lives of individuals with ASD and their families. In this thesis, I have gone beyond the evidence reported in publication 6 to give a more in depth consideration of underpinning philosophy and theory. In doing so, my analysis suggests the need for further study to understand the complex inter-related mechanisms affecting outcomes, based on a consensus theoretical framework. I have begun the process of developing an initial framework, which although not included in paper 6, is explained in summary below.

MRC guidance on complex interventions advises the development of a robust theoretical framework as the basis for good quality and robust intervention research. No consensus conceptual framework was identified in the systematic review reported; however we were able to identify factors commonly arising, which are summarised in Figure 1. These give consideration to the complex and dynamic variables affecting the context of parents prior to, during and after interventions, the mechanisms moderating parent wellbeing and outcomes measured. This preliminary framework takes account of factors drawn from the systematic review, with a focus on participation, wellbeing, the nature of ASD and the impact on families (WHO, 2007; Michie et al., 2011).

The core concept of parent participation in meaningful, pleasurable and socially expected activities is closely linked to wellbeing (Eapen, 2016), such as leisure time (Gika et al., 2012); childcare; employment and financial independence (Montes & Halterman, 2008a and 2008b). Participation is potentially affected by having a child with ASD and frameworks previously applied in relation to children (Imms et al., 2016) could equally apply to parents. Intervention outcomes are relative to pre-existing factors and may arise as a result of:

- feeling more informed, confident and skilled;
- the presence of external supports;
- the experience of feeling in control, and
- feeling supported and through participation in activities of peers and the community.

Figure 1: Proposed theoretical framework for reviewing parent focused intervention for parents of individuals with ASD over the age of 7 years



Pre-existing parent stress and QoL could influence choice of intervention and frequency of attendance (Benn et al., 2012), in turn influencing outcomes. In one study, attrition rates were attributed to wellbeing because those who dropped out, scored lower at baseline in mindfulness, personal growth and higher in stress, and anxiety (Benn et al., 2012).

In future, I would like develop this theoretical framework to underpin the development and trial of a new parent focussed intervention, based on evidence. On completion of my appraisal of research programme 3, I plan to continue to develop my practitioner and researcher skills as I apply these within the field of autism.

Before going on to discuss conclusions drawn from this thesis, I will consider strengths and limitations of the approach taken across this research.

Strengths and limitations across all studies

I have highlighted that there is a large amount of autism research but the low quality of that work and the heterogeneity of individuals included within this diagnostic umbrella, can make

it hard to draw strong conclusions. Within each publication, more specific detail is given about limitations.

Across all papers, there are limitations in the existing literature from which this work is built and this has led to the need to define concepts (e.g. how to measure wait times, what are parent focussed interventions) rather than being able to use definitions already agreed by others. Here I will summarise the strengths and limitations across the body of work, in terms of how we could improve methodologies and interpretations that can be drawn from existing and future research.

There are often on the surface, polarised views about key positions, such as whether or not we should even use diagnosis or whether interventions should be grounded in a behaviourist or social pragmatic paradigm. Through taking a pragmatist approach, the work acknowledges the complexity in the field, importance of context, intersubjectivity and shared understanding of concepts being addressed. The work provides information and a lens through which individuals, teams and local services can consider their own context. It is therefore not prescriptive, which may for some make it harder to implement, however, I would suggest that those looking for a simple answer and a quick fix will not find it. Mixed methods are recognised as adding depth and meaning, beyond simply counting wait times and allowing the generation of approaches to solve the challenges raised. I would certainly recommend this approach in future autism research. Future replication studies or studies by other researchers looking at the same questions would support the iterative process and interpretation of data in future (e.g. How effective and efficient are diagnostic assessment processes? Can these be improved using the AAA approach? What parent focussed interventions are recommended for parents of older children or adults with autism?).

The nature of the funding for this work has led to the need for a quick turn around in planning, data collection, analysis and reporting. This can have limitations in opportunities to collaborate across countries, in sample size and in time to further study each underlying concept in depth but benefits in the quick opportunities for impact of research in practice

One strength of this work, lies in its delivery through a multi-disciplinary team of practitioners and researchers and in its relevance to current issues for autistic people, their families and practitioners. In particular the involvement of services for autistic adults, adds valuable evidence in an area where there is very limited consensus currently. The work is positioned within the current research and practice context and the publications are strengthened by peer review.

A limitation has been the lack of previous research focused on wait times, service change within autism services or parent focussed interventions for older children, which means that there is yet to be consensus about some concepts related to measurement (what to measure and how to do so) or theoretical frameworks. This is perhaps most problematic within the systematic review and meta-analysis, where reliance on existing published research limits the potential to draw strong conclusions from the data. In order to account for this limitation, thorough literature review, a strong philosophical and theoretical standpoint, rigour and transparency in aims, methods, analysis and results have been central to this work.

For the wait times research, careful planning took place to identify the required sample size for the statistical analyses to be applied and a strength was success in gathering the data required. Although the work being set in Scotland could be seen as a limitation, the stratified randomised sampling approach together with reference to literature review, strengthens the transferability of results with relevance to other populations, nationally and internationally. Future studies could explicitly consider international partnerships.

Participants were professionals across different groups in the multi-disciplinary team and therefore a strength is that the results can be relevant across professions rather than being profession specific. Although all child and adult services in Scotland were approached, the group who took part in service change research, were not randomly sampled but self-selecting. This has the potential to introduce bias and therefore, in interpreting reductions in wait times, it is important to note that this occurred in services who opted to take part.

Although including practitioners who would directly apply this evidence is a strength, there is a limitation in that the views and experiences of people with autism were not sought on this occasion. This should be an important addition to future research. We currently know that the efficiency and quality of the assessment process improved using the measures applied but we do not know how this was subjectively experienced by individuals assessed.

A further limitation is the risk of bias from the research team being part of the delivery and measurement within the service change programme. The research could be strengthened in future through use of researchers completely independent of the intervention and through randomising services taking part.

In summary, as a reflective researcher it is important to be aware of the strengths and limitations of work undertaken, to inform and improve future research. I would make the following four key recommendations. Firstly, giving particular consideration to underlying concepts and theoretical frameworks and secondly introducing approaches to reduce the risk

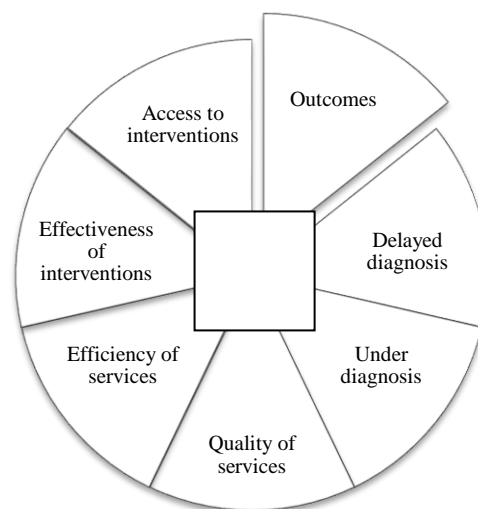
of bias through randomisation and separating those delivering change from those measuring it. Thirdly, the inclusion of the perspectives of those receiving the services is an important aspect to add validity to the interventions. Finally, it will be important to build international partnerships where there are shared challenges, interventions and the potential for collaborative work to widen the relevance of findings.

Conclusions

In presenting this compilation of my research as a PhD by published work, I have given consideration to the epistemology, theory and evidence underpinning approaches to improving the life experience of individuals with ASD and their families, either through the experience of assessment and diagnosis or through provision of evidence based supports and interventions. I have explored important underlying assumptions and demonstrated the reasons for my interest in developing a deeper and broader understanding of the ways in which key elements of service provision inter-related to jointly contribute to outcomes for individuals with ASD and their families (Figure 2).

Figure 2

Inter-related service factors affecting outcomes for individuals with ASD and their families



The international challenge of delayed diagnosis and how people with autism can access timely assessment and diagnosis was previously an area, which provided practitioners little evidence to draw from. I propose that I have made a significant contribution to this evidence base, with some indication of the relevance of this research to practice emerging, to support those designing and evaluating efficient and effective provision for children and adults. The

pragmatist philosophy and mixed methods applied to this problem, in the body of research summarised here, focussed on this challenge through considering knowledge in context and testing knowledge through empirical enquiry in a manner, which allows a more complete story to be told. The research not only gathered data about waiting times but sought to understand the challenges and solutions through consulting with practitioners. It then went further, to apply implementation science methods to seek to apply proposed solutions in practice and to evaluate their effectiveness.

My research aims and questions were driven by my developing awareness of gaps in published evidence to support effective practice, together with my pragmatist position. Additionally, they were driven by the opportunities presented by mixed methods and new theoretical perspectives, in areas with strong relevance for multi-disciplinary clinical diagnostic services and intervention provision. I have critically reflected upon the shared understanding of a phenomenon under study from interconnected objective and subjective measures. Collective actions and practice which emerge from this shared view of knowledge, contribute to the attractiveness of this approach to the pragmatist practitioner- researcher with a determination to make links between “the truth” and evidence base from research and the useful application of this in the real world.

This was demonstrated through the successful publication of the work but also in the adoption of the research in practice reported in published work and reference to our work in international clinical guidelines. Since the completion of this appraisal, I was approached by the Scottish Government and offered further grant to support implementation of our research across Scotland, with some of the direct and translational materials developed from research being made available on our website www.thirdspace.scot. Future national and international dissemination is planned but contacts from a range of interested parties suggest that aspects of diffusion of innovation have their own life undirected by the researchers.

For individuals with ASD and their families, key elements of service provision are inter-related and they jointly contribute to outcomes. Unfortunately, traditionally reported outcomes are poor and research which includes older children and adults with ASD is lacking. Despite limitations in the evidence available and in the way outcomes have been conceptualised until recently, this research portfolio presented for PhD has drawn on available frameworks and research methods, which support the application of research evidence, to take the field forward in providing creative approaches to real world research. Through providing the lens of parent ‘wellbeing’, to the consideration of ASD intervention outcomes, I have provided an extensive

and thorough systematic review and delivered evidence which can be used as the basis of a theoretical framework in future study of complex parent focussed interventions.

I have shared my aspiration to continue to develop my research in the area of interventions, with a particular interest in the challenges which arise in developing research methods in keeping with a pragmatist epistemology. There is a need to develop methods and tools which are relevant to evaluating individualised, contextually adapted approaches implemented in naturally occurring environments, as opposed to traditional ‘clinical extraction’ therapies or ‘one size fits all’ interventions and measures.

References and appendices

The appendices, which follow the references, complete the evidence submitted for this PhD by publication, with abstracts from each paper, co-author statements, curriculum vitae, a summarised self evaluation of my contribution to this published work and finally I have included each of the publications in full.

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APPENDICES

Appendix 1: Abstracts from selected publications

Paper 1

Rutherford, M., McKenzie, K., Johnson, T., Catchpole, C., O'Hare, A., McClure, I., ... & Murray, A. (2016a). Gender ratio in a clinical population sample, age of diagnosis and duration of assessment in children and adults with autism spectrum disorder. *Autism*, 20(5), 628-634.

Abstract

This article reports on gender ratio, age of diagnosis and the duration of assessment procedures in autism spectrum disorder diagnosis in a national study which included all types of clinical services for children and adults. Findings are reported from a retrospective case note analysis undertaken with a representative sample of 150 Scottish children and adults recently diagnosed with autism spectrum disorder. The study reports key findings that the gender ratio in this consecutively referred cohort is lower than anticipated in some age groups and reduces with increasing age. The gender ratio in children, together with the significant difference in the mean age of referral and diagnosis for girls compared to boys, adds evidence of delayed recognition of autism spectrum disorder in younger girls. There was no significant difference in duration of assessment for males and females suggesting that delays in diagnosis of females occur prior to referral for assessment. Implications for practice and research are considered.

Paper 2

McKenzie, K., Forsyth, K., O'Hare, A., McClure, I., Rutherford, M., Murray, A., & Irvine, L. (2015). Factors influencing waiting times for diagnosis of Autism Spectrum Disorder in children and adults. *Research in developmental disabilities, 45*, 300-306.

Abstract

Objectives: To identify the most important factors predicting delays in diagnosis for Autism Spectrum Disorder (ASD) at three stages in the diagnostic process.

Design: Cross-sectional, retrospective case notes audit

Setting: 16 (8 child and 8 adult) representative diagnosing services across Scotland.

Participants: Data from 150 case notes (80 child cases and 70 adult cases) which met the inclusion criteria that the individual had received a diagnosis of ASD and had been diagnosed by the participating service within the past 24 months.

Main outcome measures: Waiting times for ASD diagnosis at three time points: wait for first appointment; assessment duration, and total wait for diagnosis.

Results: We identified greater availability of information prior to assessment for ASD as an important factor in reducing the duration of the diagnostic process for children. Its association with diagnostic assessment duration was partly mediated by a reduction in the number of contacts required for diagnosis. In adults, having a higher a priori risk of ASD reduced the wait time from referral to first appointment, but increased the overall duration of the diagnostic process. The association between a priori ASD risk and assessment duration was partly mediated by an increase in the number of contacts required for diagnosis.

Conclusions: Within children's services, increasing the amount of relevant information available pre-assessment is likely to reduce total duration of the assessment process by reducing number of contacts required. Having a high risk of ASD as an adult, appears to result in being seen more quickly following referral, but it also appears to increase the number of contacts needed which increases assessment duration. The overall effect of this is that the total duration is longer for adults at higher risk of ASD, perhaps through increased clinical complexity.

Paper 3

Rutherford, M., McKenzie, K., Forsyth, K., McCartney, D., O'Hare, A., McClure, I., & Irvine, L. (2016c). Why are they waiting? Exploring professional perspectives and developing solutions to delayed diagnosis of autism spectrum disorder in adults and children. *Research in Autism Spectrum Disorders, 31*, 53-65.

Abstract

Background: This paper reports on the development of child and adult Action Plans for Autism Spectrum Disorder to address the problem of delayed diagnosis and lengthy waiting times. Evidence used in the development of action plans was gathered from a sequential mixed methods study to further understand the reasons for the long waiting time and potential solutions. This is the first published investigation, from the perspective of diagnosing professional teams, of the reasons for delays, which also generates solutions.

Methods: Ninety five clinicians from 8 child and 8 adult services attended 16 focus groups to explore clinicians' views on a) reducing the wait for diagnosis and b) providing a good quality diagnostic process with good adherence to clinical guidelines. During focus groups, quantitative data were fed back, used to frame discussions and facilitate solution focused action planning with each service. Sixteen local action plans were synthesised to create an ASD Action Plan for children and an ASD Action Plan for adults.

Results: Key solutions are proposed to support the reduction of the wait for diagnostic assessment, through reducing non-attendance rates, reducing inappropriate referrals, developing efficient working and communication and improving the effectiveness of care pathways. These are presented in actions plans for use by clinical teams.

Conclusion: The first step in addressing the clinical challenge of increased wait for diagnostic assessment of ASD is understanding the complex and multi-factorial reasons for delays. The action plans developed here through systematic enquiry and synthesis may provide clinical diagnostic teams with evidence based guidance on common challenges and solutions to guide future quality improvement programmes. Future research to evaluate whether using Action Plans leads to a reduction in waiting times would be of value.

Paper 4

Rutherford, M., Forsyth, K., McKenzie, K., McClure, I., Murray, A., McCartney, D., ... & O'Hare, A. (2018a). Implementation of a practice development model to reduce the wait for Autism Spectrum diagnosis in adults. *Journal of autism and developmental disorders*, 1-15.

Abstract

This study examined waiting times for diagnostic assessment of Autism Spectrum Disorder in 11 adult services, prior to and following the implementation of a 12 month change program. Methods to support change are reported and a multi-level modelling approach determined the effect of the change program on overall wait times. Results were statistically significant ($b = -0.25$, $t(136) = -2.88$, $p = 0.005$). The average time individuals waited for diagnosis across all services reduced from 149.4 days prior to the change program and 119.5 days after it, with an average reduction of 29.9 days overall. This innovative intervention provides a promising framework for service improvement to reduce the wait for diagnostic assessment of ASD in adults across the range of spectrum presentations.

Keywords

ASD, Adults, Diagnostic Assessment, Reducing wait times, Service improvement.

Paper 5

Rutherford, M., Burns, M., Gray, D., Bremner, L., Clegg, S., Russell, L., ... & O'Hare, A. (2018b). Improving Efficiency and Quality of the Children's ASD Diagnostic Pathway: Lessons Learned from Practice. *Journal of autism and developmental disorders*, 48(5), 1579-1595.

Abstract

The 'autism diagnosis crisis' and long waiting times for assessment are as yet unresolved, leading to undue stress and limiting access to effective support. There is therefore a significant need for evidence to support practitioners in the development of efficient services, delivering acceptable waiting times and effectively meeting guideline standards. This study reports statistically significant reductions in waiting times for autism diagnostic assessment following a children's health service improvement programme. The average wait between referral and first appointment reduced from 14.2 to 10.4 weeks ($t(21) = 4.3$, $p < 0.05$) and between referral and diagnosis shared, reduced from 270 to 122.5 days, ($t(20) = 5.5$, $p < 0.05$). The proportion of girls identified increased from 5.6 to 2.7:1. Methods reported include: local improvement action planning; evidence based pathways; systematic clinical data gathering and a training plan. This is a highly significant finding for many health services wrestling with the challenges of demand and capacity for autism diagnosis and assessment.

Key words

Waiting times, Autism diagnostic assessment, Children, Pathways

Paper 6

Rutherford, M. (submitted): Parent focused interventions for older children or adults with ASD and parent wellbeing outcomes: A systematic review with meta-analysis

Abstract

There is a need for better evidence in relation to parent-focussed interventions for older children (over 7 years) and adults, recommended in clinical guidelines. We conducted a systematic review of studies published between 2006 and 2016 investigating wellbeing outcomes of ASD parent focussed interventions via a search of electronic databases including MEDLINE, PsychINFO, CINAHL and the Cochrane database. We screened 9605 titles, 57 full text articles and abstracts were read. Two were systematic reviews and 22 were experimental intervention studies included for review. There were five Randomised Controlled Trials, three (non randomised) Controlled Trials and 14 (non randomised) cohort studies. Interventions were: Parent education and training (n=12); Mindfulness or relaxation training (n=6), Parent support groups (n=2) and Multi-component interventions (n=2). Studies reported five wellbeing outcomes: quality of life, parent stress, self-efficacy, parenting style and satisfaction. Separate meta-analyses compared each outcome, to test and estimate the summary effect shared by studies reporting each intervention. Statistically significant outcomes were obtained for reducing parent stress via mindfulness training and for improving parent style and satisfaction, through parent education. Analyses of a small number of studies indicate that parent focussed interventions could be effective in improving parent wellbeing, however further research is needed to determine optimal parent intervention models.

Key words: Autism Spectrum Disorder, quality of life, parent stress, intervention, family support

Appendix 2: Co-Author statement from Professor Kirsty Forsyth

Marion was Lead Research Practitioner for both phases of the Autism ACHIEVE Alliance research for which I was Principle Investigator (PI). The other PIs were Professor Anne O'Hare and Dr. Iain McClure. Professor Karen McKenzie was also a Lead Research Practitioner in the team. Marion was employed at Queen Margaret University prior to the start of the research and contributed to the design and conceptualisation of the studies from the outset, which in turn led to success with our grant bid. A programme of research (programme 2 below) ran from 2012-2013 (funded by Scottish Government, £250,000) followed by a further programme (programme 3), which ran from 2013-2014 (funded by Scottish Government, £250,000). On completion of this work, Marion continued to be employed part time at Queen Margaret University and led a piece of independent work focussed on systematic review and meta-analysis (programme 3). Throughout this period Marion has been employed only part time (2 days per week) and is also in a clinical lead role in the NHS (3 days per week). In each work stream, Marion had a pivotal role in the design and conceptualisation of the studies, in researching and synthesising the background literature for the projects and in providing a significant intellectual contribution to all of the papers below.

In research programme 1 and 2:

Marion's role was in:

- Leading the team in the work required to achieve Caldicott and Research Ethics permission.
- Leading the regular team meetings to maintain momentum for each project in programmes 1 and 2
- Liaising with participants in each phase of the research to support participation until completion of the study
- Data collection (qualitative and quantitative); and contribution to data analysis, as advised by statisticians
- Interpreting data and writing up findings for final reports and publications
- Critical review and editing of written publications

- Presentation of findings at national and international conferences

More specifically in programme 1:

- Designing the data collection tools for retrospective data analysis (service configuration tool; individual case note tool and writing guidance for each) and leading in the consultation about these tools with the project team and external expert advisors.
- Devising the national list of diagnosing services in Scotland which served as a sampling frame for randomized sampling in the study.
- Planning, organising and undertaking data collection with services across Scotland and ensuring consistency of approach between child and adult services research data.
- Interviewing service leads.
- Leading on the design, planning and implementation of focus groups across Scotland, including methods of data collection and analysis.

More specifically in programme 2:

- Recruiting participating adult services and maintaining on-going engagement and mentorship to support the change programme
- Planning and leading on the contact days for services
- Devising resources for services to support their understanding of the new adult ASD clinical guidelines
- Creating data collection tools; interpreting data returned weekly and providing written and verbal feedback to participants to support the change process
- Leading the implementation study in NHS Lothian children's services following the model applied in the adult services programme

Marion took a leading role in conceiving, planning and writing the papers published as a result of this work and subsequent work.

In research programme 3:

Marion led this research work to apply systematic review and meta-analysis methods to the topic of study. She identified the topic of study, devised methods for data extraction, data management and analysis and the article was predominantly written by her. Co-authors provided statistical support and made comments on the manuscript before submission, which Marion then used to prepare the final document.

Paper 1:

Rutherford, M., McKenzie, K., Johnson, T., Catchpole, C., O'Hare, A., McClure, I., ... & Murray, A. (2016a). Gender ratio in a clinical population sample, age of diagnosis and duration of assessment in children and adults with autism spectrum disorder. Autism, 20(5), 628-634.

This paper reports on the topical issue of gender in ASD. Marion had researched the literature on this subject during her Masters dissertation and made the case to the team that she was well placed to write this as her first lead author paper as she was very familiar with the relevant literature evidence and that this subject would be of wider interest in peer reviewed literature. This has been borne out by its acceptance in a relatively high impact journal and its growing citation index, only a year after publication. Karen McKenzie provided mentorship and supportive editing during the process but Marion selected the content and wrote the bulk of the paper.

Paper 2:

McKenzie, K., Forsyth, K., O'Hare, A., McClure, I., Rutherford, M., Murray, A., & Irvine, L. (2015). Factors influencing waiting times for diagnosis of Autism Spectrum Disorder in children and adults. Research in developmental disabilities, 45, 300-306.

Dr. Karen McKenzie was the first author for Papers 3; however Marion was closely involved in their preparation for publication, providing critical feedback and revisions to the manuscript. Feedback and contributions to academic content based on her extensive knowledge of the SIGN clinical guideline and ASD in children were particularly helpful. Additionally she provided in depth knowledge and contribution to: the conception of the papers; the planning and implementation of the research and the data collection and analysis. Both of these papers benefited greatly from these contributions. The guidelines did not come in readily auditable format and Marion's careful thought and planning supported decision making within the team in reaching agreement over the content of the factors being audited and interpretation of the data. Marion's clinical knowledge also came to the fore in meaningful interpretation of the quantitative findings of these analyses.

Paper 3:

Rutherford, M., McKenzie, K., Forsyth, K., McCartney, D., O'Hare, A., McClure, I., & Irvine, L. (2016c). Why are they waiting? Exploring professional perspectives and developing solutions to delayed diagnosis of autism spectrum disorder in adults and children. Research in Autism Spectrum Disorders, 31, 53-65.

This paper reports on the triangulation between quantitative and qualitative data collected during the research. The team were more sceptical about whether we could use the qualitative data collected through focus groups, for publication. However, Marion returned to the original data and undertook additional literature review. She has been able to frame, critically appraise and present a large amount of qualitative information in a clear and concise manner and again the role of the team was more limited to providing feedback and comments.

Paper 4:

Rutherford, M., Forsyth, K., McKenzie, K., McClure, I., Murray, A., McCartney, D., ... & O'Hare, A. (2018a). Implementation of a practice development model to reduce the wait for Autism Spectrum diagnosis in adults. Journal of autism and developmental disorders, 1-15.

This paper arose from the implementation phase of the AAA research, which took a novel approach to service change in ASD services. Marion contributed to the design and conceptualisation of the study; collection, management, analysis and interpretation of the data; contributed to the statistical analysis, and drafted and revised the manuscript.

Paper 5:

Rutherford, M., Burns, M., Gray, D., Bremner, L., Clegg, S., Russell, L., ... & O'Hare, A. (2018b). Improving Efficiency and Quality of the Children's ASD Diagnostic Pathway: Lessons Learned from Practice. Journal of autism and developmental disorders, 48(5), 1579-1595.

This paper reported the application of the body of AAA research across one health board, to implement a new pathway and reduce waiting times, over a 24 month period, demonstrating real world impact of the research. Marion led in the study design, data collection, management analysis and interpretation of data. She drafted the manuscript and led the team in revisions of the manuscript.

Paper 6:

Rutherford et al., submitted 2019: A systematic review and meta-analysis, providing an overview of the literature and current evidence about parent-focussed interventions in ASD

This paper builds on the theme of this PhD in giving intellectual consideration to support provided to families of children or adults with ASD, which was identified in the recent SIGN guidelines as a recommendation underpinned by limited evidence. Undertaking systematic review and meta-analysis are key skills Marion can take forward in her academic career and Marion's knowledge and skill in these approaches is evidenced in this paper. Marion has independently undertaken the bulk of the work required to see this work through to submission for publication, with support from Anusua (statistician) and Deborah (research assistant), with supervision and feedback from the other authors.

Appendix 2: Co-author statement from Professor Karen McKenzie

Dr. Karen McKenzie

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22nd July 2019

Co-author statement by Professor Karen McKenzie

Contribution to Published Work

I have had the opportunity to work closely with Marion Rutherford on the Autism ACHIEVE Alliance research for which I was a Lead Research Practitioner in the team. This project ran in two phases between 2012-2014 and Marion had a key role in the conceptualisation, methodology and implementation of the research during all phases. She was also key in developing data collection frameworks and forms for the different aspects of the study. The project resulted in a number of publications, and Marion has highlighted four of these that I have good knowledge of and can, therefore, comment on her extensive contribution to these papers. It should be noted that Marion was a key contributor to all of the papers that arose from this project as lead researcher for the children's services.

Rutherford, M., McKenzie, K., Johnson, T., Catchpole, C., O'Hare, A., McClure, I., ... & Murray, A. (2016a). Gender ratio in a clinical population sample, age of diagnosis and duration of assessment in children and adults with autism spectrum disorder. *Autism*, 20(5), 628-634.

This paper addresses the important issue of gender differences in the diagnosis of people with autism spectrum disorder and contributes data which age and gender differentially influence diagnosis. Marion took the lead on this paper, conceptualising the study, based on her previous research and clinical knowledge in this area. She played a key role in data collection, analysis and write up of the paper and brought her expertise to the paper,

particularly in terms of the clinical and research implications of the results, as outlined in the discussion.

McKenzie, K., Forsyth, K., O'Hare, A., McClure, I., Rutherford, M., Murray, A., & Irvine, L. (2015). Factors influencing waiting times for diagnosis of Autism Spectrum Disorder in children and adults. *Research in developmental disabilities, 45*, 300-306.

This quantitative paper identifies some of the factors that are associated with waiting times for diagnosis and the ways in which these differ for adults and children. As lead researchers for the child and adult services respectively, Marion and I worked closely on this paper. Marion created the data collection framework and gathered the child data. She also contributed greatly to every aspect of the paper.

Rutherford, M., McKenzie, K., Forsyth, K., McCartney, D., O'Hare, A., McClure, I., & Irvine, L. (2016). Why are they waiting? Exploring professional perspectives and developing solutions to delayed diagnosis of autism spectrum disorder in adults and children. *Research in Autism Spectrum Disorders, 31*, 53-65

This was a qualitative analysis of the views of professionals about the factors influencing waiting times. This paper added an extra dimension to the quantitative paper. Marion was the driving force behind this paper- conducting the data analysis from focus group interviews with staff working in adult and child services and write-up of the paper.

Rutherford, M., Forsyth, K., McKenzie, K., McClure, I., Murray, A., McCartney, D., ... & O'Hare, A. (2018a). Implementation of a practice development model to reduce the wait for Autism Spectrum diagnosis in adults. *Journal of autism and developmental disorders, 1-15*.

Marion again took the lead on writing up this paper, drawing together the process and results from the two phases of the project to report on the implementation and outcome of the initiative to reduce waiting times for Autism Spectrum Disorder in Scotland.

Appendix 3: Curriculum Vitae

Marion Rutherford

Academic Qualifications: BSc (Hons) Speech Pathology and Therapeutics, University of Glasgow, 1992. Postgraduate Certificate and Diploma in Autism (100 Masters Credits), Birmingham University, 2007. MSc in Autism Strathclyde university 2013 (60 Masters Credits).

Membership of Professional Bodies: Member of the Royal College of Speech and Language Therapists and Health Care Professions Council registered. Autism Adviser to the RCSLT (2018- present).

Employment History

Company Name	Job Title	Start Date	End Date
NHS Lanarkshire	<i>Speech & Language Therapist</i>	September 1992	June 1996
NHS Lothian	<i>Speech & Language Therapist</i>	July 1996	Current (PT)
Queen Margaret University	<i>Research Practitioner (P/T)</i>	September 2000	September 2002
Queen Margaret University	<i>Visiting Lecturer in Autism and SLT</i>	September 1996	June 2008
Queen Margaret University	<i>Research Practitioner secondment from NHS post (PT) to Firefly team</i>	October 2009	March 2011
Queen Margaret University	<i>Senior Research Fellow</i>	November 2011	Current

Publications for PhD portfolio:

1. McKenzie, K., Forsyth, K., O'Hare, A., McClure, I., Rutherford, M., Murray, A., & Irvine, L. (2015). Factors influencing waiting times for diagnosis of Autism Spectrum Disorder in children and adults. *Research in developmental disabilities*, 45, 300-306.
2. Rutherford, M., McKenzie, K., Johnson, T., Catchpole, C., O'Hare, A., McClure, I., ... & Murray, A. (2016). Gender ratio in a clinical population sample, age of diagnosis and duration of assessment in children and adults with autism spectrum disorder. *Autism*, 20(5), 628-634
3. Rutherford, M., McKenzie, K., Forsyth, K., McCartney, D., O'Hare, A., McClure, I., & Irvine, L. (2016). Why are they waiting? Exploring professional perspectives and developing solutions to delayed diagnosis of autism spectrum disorder in adults and children. *Research in Autism Spectrum Disorders*, 31, 53-65.
4. Rutherford, M., Forsyth, K., McKenzie, K., McClure, I., Murray, A., McCartney, D., ... & O'Hare, A. (2018a). Implementation of a practice development model to reduce the wait for Autism Spectrum diagnosis in adults. *Journal of autism and developmental disorders*, 1-15.
5. Rutherford, M., Burns, M., Gray, D., Bremner, L., Clegg, S., Russell, L., ... & O'Hare, A. (2018b). Improving Efficiency and Quality of the Children's ASD Diagnostic Pathway: Lessons Learned from Practice. *Journal of autism and developmental disorders*, 48(5), 1579-1595.
6. Rutherford, M., Singh-Roy-A., Rush, R., McCartney D., O'Hare A. & Forsyth K. (submitted). The nature and outcomes of parent focussed interventions for children and adults with ASD: A systematic review and meta-analysis

**Selected publications and conference proceedings not referred to in this portfolio
2015-2019**

Baxter, J., Rutherford, M. & Holmes, S. (2015) The Visual Support Project (VSP): an authority-wide training, accreditation and practical resource for education settings supporting inclusive practice. *Communication Matters*, 29(2), 9-13)

CIRCLE Collaboration (2016) *Inclusive learning and collaborative working ideas in practice (primary school 5-11 years)*. Manual. City of Edinburgh Council, Queen Margaret University & NHS Lothian: Edinburgh.

CIRCLE Collaboration (2016) *Inclusive learning and collaborative working ideas in practice (secondary school 12-18 years)*. Manual. City of Edinburgh Council, Queen Margaret University & NHS Lothian: Edinburgh.

Maciver, D., Rutherford, M., Arakelyan, S., Kramer, J. M., Richmond, J., Todorova, L., ... & O'Hare, A. (2019). Participation of children with disabilities in school: A realist systematic review of psychosocial and environmental factors. *PloS one*, 14(1), e0210511.

McClure, I. & Rutherford M. (2016) Scotland and Autism: chapter in the *Encyclopedia of Autism Spectrum Disorders* Editors: Fred R. Volkmar ISBN: 978-1-4614-6435-8 (Print) 978-1-4614-6435-8 (Online)

McKenzie, K., Forsyth, K., O'Hare, A., McClure, I., Rutherford, M., Murray, A., & Irvine, L. (2016a). The relationship between waiting times and 'adherence' to the Scottish Intercollegiate Guidelines Network 98 guideline in autism spectrum disorder diagnostic services in Scotland. *Autism*, 20(4), 395-401.

McKenzie, K., Rutherford, M., Forsyth, K., O'Hare, A., McClure, I., Murray, A. L., & Irvine, L. (2016b). The relation between practice that is consistent with NICE guideline 142 recommendations and waiting times within Autism Spectrum Disorder diagnostic services. *Research in Autism Spectrum Disorders*, 26, 10-15

Rutherford, M., McKenzie, K., McClure, I., Forsyth, K., O'Hare, A., McCartney, D., & Finlayson, I. (2016c). A national study to investigate the clinical use of standardised

instruments in autism spectrum disorder assessment of children and adults in Scotland.
Research in Autism Spectrum Disorders, 29, 93-100.

Rutherford, M. & Burns, M. (2018) Independent review of the Cornwall and Isles of Scilly pathway for recognising, diagnosing and supporting ASD in children under 18 years. Final Report. 21st December 2018 (unpublished)

Rutherford M., Baxter J., Grayson Z., Johnston L. & O'Hare A. (2019) Visual supports at home and in the community for individuals with autism spectrum disorders: A scoping review. *Autism*

Rutherford M., Baxter J., Johnston L., Tyagi V. & Forsyth K. (submitted) It's so much more than just timetables: The Home Visual Support Project Pilot Intervention Study for Parents of Children with Autism Spectrum Disorder

Scottish Intercollegiate Guidelines Network (SIGN) (2016) Assessment, diagnosis and interventions for autism spectrum disorders. Edinburgh: SIGN (SIGN publication no. 145)
Co-author and member of writing group

Conference presentations 2018-19

Rutherford M (2019) Scottish Autism Strategy conference, Strathclyde University 26th March 2019, Keynote address - National Autism Implementation Team: Locally Relevant Solutions

Eapen V., Hingorani K., Vargane Molnar M., Marion Rutherford M., Crncec R (2019) Symposium: "Measuring Quality of Life in Autism Spectrum Disorder: The QoLA Scale and Cross-Cultural Perspectives" Asia Pacific Autism Conference 2019.

Cebula K., Rutherford M, Lahood-Kullberg B, Baxter J, Johnston L (2019) Everyone likes an autism-friendly strategy: Non-autistic pupils' views of universal visual support in mainstream classrooms SARG Scottish Autism Research group.

Rutherford, Baxter, Grayson & Johnston (May 9-12th 2018) Visual Supports at Home for children with ASD, INSAR – International Society for Autism Research, Rotterdam, Netherlands

**Selected publications and conference proceedings not referred to in this portfolio
2010-2014**

Cameron, C., McClure, I., Forsyth K., Anne O’Hare, O’Hare, A., McKenzie, K., Rutherford, M., Catchpole, C., Rush, R., Murray, A., (2013) Adherence to Scottish Intercollegiate Guidelines Network 98, and the impact on duration of the diagnostic process. Poster Presentation. Royal College of Psychiatrists Conference, 18th – 20th September, Edinburgh, UK

Catchpole, C., Johnson, T., Forsyth, K., McClure, I., O’Hare, A., McKenzie, K., Rutherford, M., Rush, R., Murray, A., Arnold, J., & Chan, S. (2013). Waiting times for diagnosis of Autism Spectrum Disorder. Poster presentation. College of Occupational Therapy (COT) Annual Conference, 18-20 June, Glasgow.

CIRCLE Collaboration (2009) *Inclusive Education and Collaborative Working: a resource by teachers for teachers*. Manual. City of Edinburgh Council, Queen Margaret University & NHS Lothian: Edinburgh.

CIRCLE Collaboration (2011) *Up, Up and Away! Literacy in the Early Years - Identifying need*. Manual. City of Edinburgh Council & Queen Margaret University: Edinburgh.

CIRCLE Collaboration (2011) *Up, Up and Away! Literacy in the Early Years - Planning to meet the need*. Manual. City of Edinburgh Council & Queen Margaret University: Edinburgh.

Rutherford M (2011) Community Child Health Lectures, NHS Lothian. Language, Literacy and Learning: implications for Nursery and P1, Promoting Recovery for at risk children

CIRCLE Collaboration (2012) *Teachers ideas in practice: Supporting Learners needs in Secondary Schools*. Manual. City of Edinburgh Council, Queen Margaret University & NHS Lothian: Edinburgh.

Harden, J., Johnson, T., Rutherford, M., Forsyth, K., McClure, I., O'Hare, A., McKenzie, K., Catchpole, C., Rush, R., Murray, A., Arnold, J., & Chan, S. (2013) The role of the OT in the diagnosis of ASD. Poster Presentation. International Allied Health Professions Conference, 03 October, Edinburgh, UK.

Harden, J., Catchpole, C., Johnson, T., Forsyth, K., McClure, I., O'Hare, A., McKenzie, K., Rutherford, M., Rush, R., Murray, A., Arnold, J., & Chan, S. (2013). Waiting times for diagnosis of Autism Spectrum Disorder. Oral/Poster Presentation. International Allied Health Professions Conference, 03 October, Edinburgh, UK.

Johnson, T., Rutherford, M., Forsyth, K., McClure, I., O'Hare, A., McKenzie, K., Catchpole, C., Rush, R., Murray, A., Arnold, J., & Chan, S. (2013). The role of the OT in the diagnosis of ASD. Oral presentation. College of Occupational Therapy (COT) Annual Conference, 18-20 June, Glasgow.

McAlpine R, Rutherford M, Forsyth K, Maciver D & McLeod I (2012) Occupational Focussed Early Intervention: an evidence based resource to support occupational engagement. oral presentation College of Occupational Therapists conference June 2012

McClure, I., Catchpole, C., Forsyth, K., Johnson, T., McKenzie, K., O'Hare, A., Rutherford, M., Rush, R., & Murray, A. (2013). Adherence to Clinical Guidelines for ASD Diagnosis in Child and Adult Services in Scotland. Poster Presentation. SIGN 20th Anniversary event, 4th November, Edinburgh, UK.

McClure, I., O'Hare, A., McKenzie, K., Forsyth, K., & Rutherford, M. (2013). Factors influencing waiting times for diagnosis of Autism Spectrum Disorder in children. Royal College of Psychiatrists Faculty of Child and Adolescent Psychiatry Conference, 18-20th Sept 2013, Edinburgh.

McClure, I., Catchpole, C., Forsyth, K., Johnson, T., McKenzie, K., O'Hare, A., Rutherford, M., Rush, R., & Murray, A. (2013). Adherence to Clinical Guidelines for ASD Diagnosis in Child and Adult Services in Scotland. Oral/Poster Presentation. European Academy of Childhood Disability (EACD), 10 – 12 October, Newcastle, UK.

McClure, I., Catchpole, C., Forsyth, K., Johnson, T., McKenzie, K., O'Hare, A., Rutherford, M., Rush, R., & Murray, A. (2013). Adherence to Clinical Standards and Guidelines for ASD Diagnosis in Child and Adult Services in Scotland. Poster Presentation. International Meeting for Autism Research (IMFAR), 2 - 4 May, Spain.

McClure, I., Catchpole, C., Forsyth, K., Johnson, T., McKenzie, K., O'Hare, A., Rutherford, M., Rush, R., & Murray, A. Adherence to Clinical Guidelines for ASD Diagnosis in Child and Adult Services in Scotland. Action on Autism Seminar, 26/27 November 2013, University of Strathclyde, UK

McKenzie, K., Catchpole, C., Forsyth, K., Johnson, T., McClure, I., O'Hare, A., Rutherford, M., Rush, R., Murray, A. & Peter, A. Factors that influence time taken to reach ASD diagnosis in child and adult services Action on Autism Seminar, 26/27 November 2013, University of Strathclyde, UK

McKenzie, K., McClure, I., O'Hare, A., Forsyth, K., Rutherford, M., Catchpole, C., Johnson, T., Arnold, J., Chan, S. Rush, R. & Murray, A.L. (2013). Factors influencing waiting times for diagnosis of Autism Spectrum Disorders: A comparison of child and adult services. Oral presentation. British Psychological Society Annual Conference, 9-11 April, Harrogate.

McKenzie, K., Catchpole, C., Forsyth, K., Johnson, T., McClure, I., O'Hare, A., Rutherford, M., Rush, R. & Murray, A. (2013). Factors that influence time taken to reach ASD diagnosis in child and adult services. Oral/Poster Presentation. European Academy of Childhood Disability (EACD), 10 – 12 October, Newcastle, UK.

O'Hare, A. Catchpole, C., Forsyth, K., Johnson, T., McClure, I., McKenzie, K., Rutherford, M., Rush, R., & Murray, A. (2013). Using Mixed Methodology to Investigate ASD Diagnosis. Poster Presentation. International Meeting for Autism Research (IMFAR), 2 - 4 May, Spain.

O'Hare, A. Catchpole, C., Forsyth, K., Johnson, T., McClure, I., McKenzie, K., Rutherford, M., Rush, R., & Murray, A. (2013). Using Mixed Methodology to Investigate ASD Diagnosis. Oral/Poster Presentation. European Academy of Childhood Disability (EACD), 10 – 12 October, Newcastle, UK.

O'Hare, A., Catchpole, C., Forsyth, K., Johnson, T., McClure, I., McKenzie, K., Rutherford, M., Rush, R., & Murray, A. Using Mixed Methodology to Investigate ASD Diagnosis. Action on Autism, 2nd Seminar, Clinical Research, Interventions and Impact, 4/5th February 2014, University of Strathclyde, UK.

Rutherford, M., Catchpole, C., Forsyth, K., Johnson, T., McClure, I., McKenzie, K., & O'Hare, A. (2013). An Action Plan for Improving Efficiency and Quality of the Process of ASD Diagnosis in Adults and Children. Poster Presentation. International Meeting for Autism Research (IMFAR), 2 - 4 May, Spain.

Rutherford, M., Johnson, T., Forsyth, K., McClure, I., O'Hare, A., McKenzie, K., Catchpole, C., Rush, R., Murray, A., Arnold, J., & Chan, S. (2013). The use of contextual information in reducing waiting times for ASD diagnosis in children. Oral presentation. College of Occupational Therapy (COT) Annual Conference, 18-20 June, Glasgow.

Rutherford M (2012) Castang Conference 2012: Helping young children develop language in readiness for literacy. oral presentation

Rutherford M and Johnston L (2012) Community Child Health Lecture, NHS Lothian 2012: Communication Strategies for school aged children with autism. Oral presentation

Rutherford, M., Catchpole, C., Forsyth, K., Johnson, T., McClure, I., McKenzie, K., & O'Hare, A. (2013). An Action Plan to Improve Efficiency and Quality of the Process of ASD Diagnosis in Adults and Children. Oral Presentation. International Allied Health Professions Conference, 03 October, Edinburgh, UK.

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Member of SIGN guideline development group:

Scottish Intercollegiate Guidelines Network (SIGN) (2007) Assessment, diagnosis and clinical interventions for children and young people with autism spectrum disorders. Edinburgh: SIGN (SIGN publication no. 98)

Scottish Intercollegiate Guidelines Network (SIGN) (2016) Assessment, diagnosis and interventions for autism spectrum disorders. Edinburgh: SIGN (SIGN publication no. 145). [June 2016]. Available from URL: <http://www.sign.ac.uk>

Unpublished dissertations

Rutherford, M (1992). Pragmatic Skills in children with Autism. Thesis submitted for BSc (Hons) Speech Pathology and Therapeutics, Glasgow University

Rutherford, M (2013). A retrospective case note audit to compare the diagnostic pathway and characteristics of girls and boys diagnosed with an ASC (Autism Spectrum Condition). Masters Thesis submitted to University of Strathclyde, as part of the MSc in Autism

Appendix 4: Self-Evaluation of Contribution to Published Work

As a researcher at Queen Margaret University I have worked on several research projects. My interest in research initially stemmed from my undergraduate research project, which was a study of pragmatic skills in children with ASD (Rutherford, 1992). During undergraduate training, I worked with children with autism and was very aware of the lack of evidence for practitioners in relation to how to identify and diagnose ASD or how to offer effective support and intervention for the core social communication difficulties found in ASD. In my clinical work, I gained extensive experience and knowledge about ASD but continued to be challenged by the lack of evidence base for practice. When I had the opportunity to be part of the prosody research team, in developing the methodology and measurements of prosody, collecting data and interpreting data analysed, my interest in the possibility of becoming more involved in research was sparked. Additionally, I had the opportunity to be a core member of the Autism SIGN guideline development group in 2004 – 2007. This experience enhanced my skills in critical appraisal, and skills in writing to synthesise complex information and I was motivated to undertake Masters level study.

My clinical work has provided a rich source of ideas about aspects of research, which are important to clinicians, families and individuals with ASD, and I have an ongoing aspiration that the work I undertake has a positive and useful real world impact. I observed that the wait for diagnosis was very distressing to families and that it affected access to services – an observation, which was later, re-affirmed through academic study. Solutions to the long wait for ASD diagnosis were possible in some areas but not in others and I took the view that clinicians needed more evidence about what to change and how to change it.

The opportunity to be part of the CIRCLE team and lead on an early years project, was my first paid research post, which allowed for a larger part of my working week to have a research focus. This work was closely related to my clinical work with children in areas of social deprivation and although it did not have an ASD focus, there were many overlaps in the literature and evidence base, related to early typical or non-typical development of language, cognition, learning and participation.

In 2011, I took the opportunity to be a lead researcher in the Autism ACHIEVE Alliance Team in response to a government call for funding into waiting times for diagnostic assessment of ASD. As I analysed and reported on data collected from the retrospective case

note analysis about the waiting times, I formulated the hypothesis that solutions to improving waiting times could be identified through combining the data collected with qualitative research with practitioners. This mixed method approach proved to deliver rich data with real world applicability and further motivated me to lead a further research programme with clinical services to reduce waiting times for ASD assessment through a combination evidence based action plans developed in the early part of the research programme and a previously reported service change methodology.

It was at this point that I began to consider PhD by publication and in considering the evidence for the value of early or timely diagnosis, I became interested in why this was so important and its influence on the outcomes of individuals diagnosed with ASD. An HTA study into outcome measures in ASD (McConachie et al., 2015) highlighted the growing change in focus from impairment focussed to participation focussed outcomes and identified the need for more intervention studies to measure quality of life and family stress. I was invited again, to be part of the SIGN ASD guideline development and writing group (2014-2016). Through this work, the limited evidence related to interventions to address these outcomes, became increasingly apparent to me. I would like to further develop my research interests in this area. In order to do this, I have undertaken a systematic review and meta-analysis of outcomes from parent focussed ASD interventions in children and adults as a basis for seeking future research grants.

In my clinical – researcher role, between 2014-2019 I also led the development of resources for schools and parents in relation to ‘visual supports’ – a key evidence based intervention to support communication and participation of individuals with ASD. I have a growing number of publications related to this area of work, cited in my CV. These provide evidence of the breadth of my work, my increasing confidence and my independence in working in research at a doctoral level.

In 2019, I accepted an invitation by the Scottish Government Autism Strategy team to lead a new ‘National Autism Implementation Team’, with grant funding, focussed on continued implementation of research into practice. The aim of this team of making and measuring real world differences to individuals with autism across the lifespan and their families, through work on: Assessment and Diagnosis Pathways; Supporting individuals with autism in schools and their teachers; Employment and Community engagement. I anticipate that this

role will give me further opportunities to advance my academic career in this field, whilst grounded in real world contexts.

Thus, my critical appraisal can draw on a broad but coherent and ongoing track-record. What follows is a description of my journey as a practitioner- researcher to date via selected publications I have selected for this doctoral portfolio, with indicative citation counts from Google Scholar (26.08.19).

1. **Rutherford, M., McKenzie, K., Johnson, T., Catchpole, C., O’Hare, A., McClure, I., ... & Murray, A. (2016). Gender ratio in a clinical population sample, age of diagnosis and duration of assessment in children and adults with autism spectrum disorder. *Autism*, 20(5), 628-634.**

My contribution: This paper reports key findings that the gender ratio in this consecutively referred cohort is lower than anticipated in some age groups and reduces with increasing age. The gender ratio in children, together with the significant difference in the mean age of referral and diagnosis for girls compared to boys, adds evidence of delayed recognition of autism spectrum disorder in younger girls. For this paper I jointly collected and analysed the data and I was responsible for the writing and generation of ideas in the discussion

Impact Factor: 3.6

Citations: (n = 54) 26.08.19

This paper was the first research report, for which I was lead author. The idea for the paper came from my Master’s Thesis dissertation, in which I had researched the gender differences in the children attending my own clinic for ASD diagnostic assessment. I was aware that gender differences in ASD are very topical and the data we had collected provided an ideal opportunity for further analysis, which added to the relatively limited evidence available in this field. This was reflected in it being accepted in an autism journal with a relatively high impact factor.

2. McKenzie, K., Forsyth, K., O'Hare, A., McClure, I., Rutherford, M., Murray, A., & Irvine, L. (2015). Factors influencing waiting times for diagnosis of Autism Spectrum Disorder in children and adults. *Research in developmental disabilities*, 45, 300-306.

My contribution: This paper presents key quantitative findings from the regression analysis of the data collected from the retrospective case note analysis. In this project there were two leads – one for child and one for adult services. As lead researcher for the child services, I created the tools used for data collection and collected the child service data personally. I oversaw the data entry and data cleaning in preparation for the regression analysis. For this paper, Karen McKenzie and Aja Murray performed the analyses and prepared the written draft of the publication. I contributed ideas in the planning, methods and discussion. I provided review and editing prior to submission.

Impact Factor: 1.9

Citations: (n = 9) 26.08.19

This paper was the first paper written by the team and was based on the final project report. I was an equal partner in developing the research concepts and study aims, which were the basis for the project plan. I undertook the practical aspects of the work to collect collate and interpret data as well as writing the project report for the Scottish Government. At this point, I was a novice in writing for publication and benefited greatly from the opportunity to do this jointly with experienced colleagues. The expertise in regression analysis lay with my colleague and in working jointly, I learned about this method, as well as ways of planning and writing a paper for publication.

- Rutherford, M., McKenzie, K., Forsyth, K., McCartney, D., O'Hare, A., McClure, I., & Irvine, L. (2016). Why are they waiting? Exploring professional perspectives and developing solutions to delayed diagnosis of autism spectrum disorder in adults and children. *Research in Autism Spectrum Disorders, 31*, 53-65**

My contribution: For this paper, as well as conceiving and writing the paper, I had taken the lead in the planning, delivery and analysis of the data collected from focus groups in which we used a sequential mixed method design, where the researchers fed back quantitative results from the casenote analysis to each service prior to each group. Each group used solution focussed planning to develop local action plans. Karen and I jointly worked to synthesise the data to develop the aggregated action plans for child and adult services.

Impact Factor: 1.9

Citations: (n = 9) 26.08.19

This paper is very important, in this body of work, which uses innovative methods to provide novel insights into evidence based solutions for clinical practice. This paper was a labour of love and one I was mainly responsible for within the team from the outset. It presents the qualitative element of the work done which was an area I was more comfortable and familiar with, having used focus group methodology and thematic analysis in the past. As a clinician and researcher, I was highly motivated by this opportunity to present the nuances and real world application of our findings alongside the quantitative data. There are very few qualitative studies published in this field and I was very pleased at the publication being accepted. The evidence shared in this paper was pivotal to the development of the research, which led to papers 4 and 5 below and is evidence of the way in which the separate phases of research are closely linked.

4. **Rutherford, M., Forsyth, K., McKenzie, K., McClure, I., Murray, A., McCartney, D., ... & O'Hare, A. (2018a). Implementation of a practice development model to reduce the wait for Autism Spectrum diagnosis in adults. *Journal of autism and developmental disorders*, 1-15.**

My contribution: This paper builds on the previous work and publications, presents new data and brings together the expertise of Kirsty Forsyth in evidence based approaches to service change, and research skills and ASD knowledge within the wider team, with my own expertise and knowledge of ASD research evidence. Having built up experience as a researcher, I was able to take the lead in undertaking this research and contributed to the planning, delivery of the intervention, data collection, data analysis and writing up in relation to the changes services could make that would effect change, developed from my research over the preceding 2-3 years represented in the publications from 2015 and 2016.

Impact Factor: 3.3

Citations: (n=1) 26.08.19

This research gave me the opportunity to use another set of research skills, broadening my portfolio of research experience. The type of research collaboration achieved in this work was very powerful and led me to engage in a much deeper level of understanding of theoretical underpinnings of practice development, moving from the position of the least experienced member of the team to a position of confidence to lead and manage the project, whilst ensuring rigour, quality and timely outcomes.

5. **Rutherford, M., Burns, M., Gray, D., Bremner, L., Clegg, S., Russell, L., ... & O'Hare, A. (2018b). Improving Efficiency and Quality of the Children's ASD Diagnostic Pathway: Lessons Learned from Practice. *Journal of autism and developmental disorders*, 48(5), 1579-1595.**

My contribution: This paper builds on the previous work and publications and presents new data highlighting the real world impact of this body of work. Having built up experience as a researcher I was able to take the lead in undertaking this research and contributed to the planning, delivery of the intervention, data collection, data analysis and writing up in relation to the changes within children's services to reduce waiting times

Impact Factor: 3.3

Citations: (n = 4) 26.08.19

In this work, my co-authors were all clinical experts who were less motivated by data and more motivated by making effective service changes. I was able to lead on this work and to apply research skills in my own clinical context, where measurement of impact is not often easy to do. However the results, which led to a statistically significant reduction in wait times across our region, became key to future service planning and funding as it provided evidence of factors affecting demand and capacity within a clinical service and supported clinicians to evidence that their 'small changes' had a big effect. I have been pleased to hear that this publication, alongside other Autism ACHIEVE Alliance publications has been used by other clinical services across the UK as a basis for planning service improvement.

6. **Rutherford, M. (Submitted) The nature and outcomes of parent focussed interventions for children and adults with ASD: A systematic review and meta-analysis**

My contribution: This paper is a critical review of the literature. I was responsible for proposing, framing and planning the evaluation; undertaking an appropriate systematic literature review and appraisal, following PRISMA guidelines; identifying studies for meta-analysis and interpreting findings as well as reporting findings, writing the publication and drawing together recommendations for future research.

Impact Factor: (n/a)

Citations: (n = n/a)

This paper was the most recently written and *submitted* paper which builds on work from my early research career. My work in this field has led me to undertake a number of literature reviews, however, in order to meet the requirements for publication I have expanded on this significantly in quantity and quality. As this is a critical review, it is similar to the literature review in a traditional PhD, showing a critical and detailed knowledge of a substantial body of research. I was responsible for searching for appropriate literature and critically appraising it, drawing together recommendations for future research. I will bear these recommendations in mind for our own research. Writing this paper gave me the opportunity to become fully confident using search strategies and databases such as Medline, a skill which is invaluable in a research career.

Writing for publication during the last 3 years has given me the opportunity to improve my writing skills and gain confidence in communicating with my academic peers. This critical review has supported my development of a deep knowledge and understanding of the literature in my specialist area, providing a critical overview of the subject for both myself and my peers, using “gold standard” methodologies of systematic review and meta-analysis.