A Rapid Realist Review of diagnostic assessment pathways for children and young people with possible Autism.

RE-ASCeD: A Realist Evaluation of Autism ServiCe Delivery

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ADHD Attention deficit hyperactivity disorder

ADOS Autism Diagnostic Observation Schedule

AHP Allied Health Professional

ASC Autism Spectrum Condition

ASD Autism Spectrum Disorder

BACCH British Association of Community Child Health

BACD British Academy of Childhood Disability

CAMHS Child and Adolescent Mental Health Services

CCG Clinical Commissioning Group

CCH Community Child Health

CDC Child Development Centre

CPD Continuing Professional Development

CDS Child Development Services

CDT Child Development Team

CMO Context, Mechanisms, Outcomes

CQC Care Quality Commission

CRN Clinical Research Network

CYP Children & young people

GDA General Developmental Assessment

GP General Practitioner

InCo Area Inclusion Co-ordinator

M-CHAT Modified Checklist for Autism in Toddlers

MDT Multi-disciplinary team

NDC Neuro-developmental conditions – see Will comment re NDD

NHS National Health Service

NHSE National Health Service England

NICE National Institute for Health and Care Excellence

NIHR National Institute for Health Research

NPT Normalisation Process Theory

PPI Patient and Public Involvement

PT	Programme Theory
QI	Quality Improvement
RASDN	Regional Autistic Spectrum Disorder Network
RCPCH	Royal College of Paediatrics and Child Health (UK)
RCPsych	Royal College of Psychiatrists
RE	Realist Evaluation
RRR	Rapid Realist Review
SALT	Speech and language therapist
SENCO	Special Educational Needs Co-ordinator
SIGN	Scottish Intercollegiate Guidelines Network
STAT	Screening Tool for Autism in Toddlers and Young Children
UK	United Kingdom of Great Britain and Northern Ireland
WP	Work Package

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Lay Summary

How can services that assess children for autism make accurate and quick decisions?

Autism is a lifelong condition, with symptoms usually emerging during the first two years of life and persisting throughout an individual's life. People can receive a diagnosis at any age. Parents often notice signs that something may not be quite right, but it might also be a teacher, doctor, or someone else. When this happens, the child and their parent see a specialised diagnosis team.

Diagnosing autism is not easy. It involves gathering information about a person's behaviour, functioning, and development by more than one type of professional. Another reason is that professionals might assess a child for other conditions too. The number of children referred for assessment has been gradually increasing over the last few years. As a result, many families spend more than a year waiting for a diagnosis.

In 2019, the NHS made a commitment for more families to get the right help, quicker. To do this, waiting times need to be shorter so children will get the right support sooner. This project aims to guide the people who plan services for autistic children and their families.

This is the first of four stages in a bigger research project. This stage looked for any evidence from the UK that discussed how to work out if a child or young person may have autism. We discussed the evidence with the project team which included groups/people who support autistic people. We found there are many different approaches to diagnosis, for example, with respect to:

- which professional makes the diagnosis,
- how many professionals are involved,
- what tests are carried out.

We also found that families wanted to have their say, know what is going to happen and how long things will take. Families liked having good support and information before, during and after their child was (or was not) diagnosed. Parents liked to have a report that said what their child was good at, not just what they found difficult. They also found it helpful if the report suggested how to help their child.

Professionals wanted to have more training to understand autism. Sometimes they found it complicated to know which specialist should assess a child or young person and this caused delays. Most professionals understood that it can be upsetting for some parents to be told that their child is on the autism spectrum. Professionals also knew that often this was because local post-diagnostic support services were few and far between.

This review of the current evidence will be used as a basis for the next stages of the project:

- A national survey of Autism Diagnostic Services to see what is currently happening in diagnostic services
- A more detailed look at some examples of services from around England. This will include interviews and focus groups with autistic children and their families, and the professionals involved in their local autism services.

EXECUTIVE SUMMARY

Background

Autism is characterized by the presence of persistent deficits in the ability to initiate and sustain reciprocal social interaction and social communication, and by a range of restricted, repetitive, inflexible patterns of behaviour and interests. Presentation varies significantly in relation to severity and co-occurring intellectual, neurodevelopmental or mental health disorders. Diagnostic assessment of children and young people (CYP) with possible autism is usually carried out by services based in either Child Development Services/Paediatrics (CDSs) or Child and Adolescent Mental Health Services (CAMHS), or tertiary services. However, the approach to diagnostic assessment varies considerably around the UK and families frequently report dissatisfaction with the process, particularly long waits from referral to diagnosis.

This Rapid Realist Review (RRR) of the current evidence is the first step in a national **R**ealist **E**valuation of **A**utism **S**ervi**Ce** **D**elivery (RE-ASCeD) and explores how particular approaches may deliver high quality and timely autism diagnostic services for CYP with possible autism. Research questions for the RRR are as follows:

1. How do various models of autism diagnostic and support services address the differing needs of different service user groups and what contexts and mechanisms affect their ability to do so?
2. How do models of autism diagnostic and support services improve service user diagnostic experience?
3. What aspects of implementation, staffing and organisational context influence how models of autism diagnostic and support services operate?

Method

The complexity of autism diagnostic pathways in terms of differences in local service configurations and settings, lends itself to a realist review method that can tease out contextual factors, resources, and responses of those delivering and accessing the services. Realist reviews present evidence as programme theories, or key features of the service with a description of what appears to be leading to certain outcomes. These programme theories are supported by details of the context (C), mechanisms (resources and responses of those delivering and accessing the service) (M), and the intended (and unintended) outcomes (O). These are presented as CMO configurations. RRR is a well-established approach to synthesising evidence within a compressed timeframe to identify models of

service delivery leading to desired outcomes. This study included the iterative five stages of RRR: developing and refining the scope of the review; searching and identifying information; extracting and appraising the evidence; synthesizing information; and interpreting and refining programme theories.

With the support of our Expert Stakeholder Group, we collected 129 grey literature and policy and guidelines papers from the background search, and 211 articles from the primary search. Based on the iterative screening results, data extraction was carried out using a hybrid approach: basic details from each included article (n=79) were recorded in a data extraction form in Excel; highly relevant articles (n=36) were coded in NVivo. Based on the analysis of individual papers and cross-evidence comparison (36 from primary and background search and 9 from iterative secondary search, see Appendix B), we then built programme theories and refined CMO configurations. A workshop with our Expert Stakeholder Group helped test and refine our programme theories adding to the rigour of our findings.

Findings

We developed seven programme theories in collaboration with our Expert Stakeholder Group. The first four focus on the referral pathway and the last three relate to issues that run through the whole pathway. In brief:

1. **Recognition**: **Parents/carers concerns are listened to and frontline professionals are cognisant of Autism and referral pathways.**
    - Parents sometimes felt that frontline health and education professionals did not listen to their concerns or refer their child to the appropriate service early enough, and perceived this to cause significant delays in diagnostic assessment.
    - Professionals were not always confident in recognising the signs and symptoms of autism or cognisant with referral pathways; they also had to strike a balance between taking parents' concerns seriously and referring in a timely manner to the appropriate service.
    - Greater autism awareness and training for frontline professionals, particularly GPs and teachers, alongside training in how, when and who to refer to may facilitate appropriate and timely referral and reduce parental frustration.
2. **Referral process and triage: referrals provide relevant information to enable timely triaging.**
    - Referrals often lacked relevant information. If autism diagnostic services provide clear guidelines for referrers on what information is needed and how to provide it (e.g. a proforma), this can save time at the triaging stage.

- Cross-organisational triaging, or a single point of access, avoids referrals bouncing between agencies and promotes integrated working.
- When declining a referral, an explanation should be provided.
- These measures may reduce the waiting list and low diagnostic yield (low numbers of positive diagnoses).

3. **Diagnostic model**: **skills mix, clinical judgement and digital technology**
   - There were many different models for autism diagnostic services and no definitive evidence to support one over another.
   - However, core multi-disciplinary teams are advisable, with access to other disciplines including child psychiatry, and an assets-based approach, focusing on a CYP's strengths as well as difficulties. This is in line with the approach recommended in the NICE Guidelines (2011) and Quality Standard (2014).
   - Approaches to manage staff shortages (e.g. paediatricians and child psychiatrists) and to streamline the assessment process included a structured and consistent approach, extending the role of available staff (e.g. speech therapists) and developing efficient working and communication practices.
   - Clinicians' experience, expertise and confidence in their diagnostic skills appeared to influence the balance of interview, observation and recognised tools.
   - Digital technology, for example using computers at school to collect observational data, is in an early stage and there is little data on whether this leads to a valid diagnosis. Similarly, remote observations and consultations (in the context of covid-19) appear to have largely positive feedback from parents and CYP but this stems from necessity rather than diagnostic validity, confidence in findings and relationship with the clinical team.
4. **Diagnostic feedback: understanding feedback and developing a management plan**
   - Parents found a positive diagnosis distressing, even when anticipated, and found it hard to absorb information during the feedback session.

     Parents valued various strategies including a structured approach to feedback; opportunity to ask questions; a further appointment for more discussion; and clinicians using straightforward terminology and responding sensitively.
   - Reports should be timely and in a format that everyone finds helpful. They should include the assessment and diagnosis outcome and a tailored intervention care plan, co-developed with parents and taking an assets-based approach.

- The report should also include which interventions will be offered, taking account of co-existing conditions; what support will be available; information about relevant agencies; and contact details for a 'link person'.

5. **Working in partnership with families: parents as co-experts**
   - When parents understood the diagnostic process in advance, including reasons why it takes the time it does, this improved their satisfaction and acted to moderate expectations.
   - However, the diagnostic pathway is stressful so parents found it helpful to have a single point of contact; to be provided with timely explanations; and included in decision-making. These strategies also increased parental engagement with services.
   - This highlights the importance of integrating the perspectives of CYP, their family, and professionals to ensure the quality of the process.
   - Parents valued open, honest dialogue - by co-creating a relationship with families, clinicians can assist them to develop strategies to manage the difficulties they are likely to encounter.
6. **Inter-agency working**: **policy, funding, inter-disciplinary and cross agency working**
   - Despite the policy drive for integrated services (macro level) there appears to have been limited progress, for example CAMHS and CDS often sit in different NHS Trusts (meso level), each commissioned to deliver specific pathways.
   - However, integrating the pathways into a single assessment process potentially saves time, costs less and the time taken to reach diagnosis may be reduced dependent on factors such as case complexity.
   - Most guidelines refer to MDTs as best practice for autism diagnostic assessment and follow-up but lack recommendations about how roles within (or across) teams are negotiated and how disagreement is resolved. Where teams worked well together, it appeared to be based on good personal relationships (micro level).
   - However, there was limited evidence of health and education working together and parents took on the role of coordinating services. Where available, an Additional Learning (or Special Educational) Needs Co-ordinator provided the link between parents, teachers and other professionals.
   - If all stakeholders work in partnership and the knowledge generated is effectively embedded into local services, this will build capacity, improve parent/CYP satisfaction and support planning of services both locally and nationally.
7. **Training, service evaluation and development: professionals working with CYP in community and specialist settings**

- Several papers identify the importance of effective training in improving the quality and efficiency of autism diagnostic services. Training would benefit from a clear framework that identifies what skills or competencies are required depending on the level of involvement and likely impact of contact with individuals with autism.
- However, autism awareness training is relevant to all professionals (health, social care and education) who work in settings where they will come across CYP with autism.
- Multi-agency training for professionals with a targeted and coordinated approach across organisations can facilitate a wide breadth of basic training.
- Finally, Continuing Professional Development should be used to review service development, for example, investigating the utility of new diagnostic measures, and services should maintain or develop efficient systems of collecting information about referrals, waiting times and outcomes to support audit and self-evaluation.

Conclusions

To our knowledge this is the first theory informed review of the diagnostic pathway for CYP with possible autism in the UK. Many of the issues identified in our review would be addressed by full adherence to NICE guidelines and quality standards (2011, 2014) but we identified seven key areas for further exploration, stemming from the programme theories. The wider Re-ASCed study will provide the opportunity to explore some of these areas, depending on the case studies selected. The areas are:

1. Evaluation of training and support materials available for a) non-specialist staff and b) parents and CYP accessing the service.
2. Evaluation of training packages for those working in autism services to upskill, and evaluation of the impact on workforce shortages.
3. Evaluation of asset-based approaches to diagnosis and support in autism.
4. Evaluation of the barriers and facilitators to comprehensive needs-led diagnostic assessment.
5. Evaluation of approaches to integrating services dealing with autism.
6. Evaluation of the use of digital technology in diagnostic assessment.

1: BACKGROUND AND RATIONALE

1.1 Autism: an overview

Autism Spectrum Disorder (to use the current diagnostic coding term), or Autism Spectrum Condition to use the term many families prefer is characterized by the presence of 'persistent deficits in the ability to initiate and sustain reciprocal social interaction and social communication, and by a range of restricted, repetitive, inflexible patterns of behaviour and interests' (American Psychiatric Association, 2013). Autism affects around 1-2% of children and young people (CYP) and the prevalence appears relatively consistent globally (Baird et al., 2006, Elsabbagh et al., 2012, Waugh, 2017, Baio et al., 2018). We will use the term 'autism' because it has been endorsed by autistic adults, family members/friends and parents (Kenny et al., 2016).

Co-morbid mental health disorders are common, with elevated rates of autism found within clinical populations, for example within Child and Adolescent Mental Health Services (CAMHS) (Simonoff et al., 2008, Wistow and Barnes, 2009, Joshi et al., 2010). Presentation varies significantly in relation to severity of these deficits, intellectual ability or disability and the child's verbal abilities (American Psychiatric Association, 2013). Many children will experience significant levels of disability, including co-existing conditions, behaviour that challenges and secondary mental health difficulties such as Anxiety Disorder and Depression. Individuals with autism, whether with associated learning difficulties or not, contribute significantly to in-patient Tier 4 CAMHS admissions, particularly in girls without learning disability, often only receiving a diagnosis once in that setting (Health and Social Care Information Centre, 2018). Similarly, a recent survey of adults who have attempted suicide, identified a high prevalence of previously unrecognised autistic traits, particularly in women (Richards et al., 2019).

People with autism may depend on adult support and/or care throughout their lives and the economic effects of autism in those with or without a learning difficulty can be considerable, in particular that of special education (including early intervention services), and indirect costs, such as parental productivity loss (Buescher et al., 2014). In young children with autism, higher costs are associated with increasing age and symptom severity (Knapp et al., 2007, Barrett et al., 2012). The impact of autism and behavioural difficulties on families can be considerable, with parents of children with autism experiencing higher levels of stress than parents of children with other disabilities (Hayes and Watson, 2013). Petrou et al. (2018) suggest this is particularly so where the child has coexisting conditions, sleep problems, communicated only by physical means, or had severe autistic symptomatology.

1.2 The policy context: NICE guidelines

The NHS Long Term Plan (NHS England, 2019a, p.53) acknowledges that CYP with possible autism wait too long for diagnostic assessment and aspires to 'implement the most effective ways to reduce waiting times for specialist services… achieving timely diagnostic assessments in line with best practice guidelines'. The need to improve the diagnostic pathway for CYP with possible autism has been widely acknowledged (Reed and Osborne, 2012, Crane et al., 2016, Rutherford et al., 2018), including a recent campaign by the National Autistic society (2019); professional bodies such as the British Association of Community Child Health (BACCH) and British Academy of Childhood Disability (BACD) (Male and Reddy, 2018); lobbying groups including Autistica and the wider coalition to which it belongs, Embracing Complexity; and the James Lind Alliance (2016) priority setting exercise for autism.

In the UK, NHS diagnostic practice for CYP with possible autism is based on National Institute for Heath and Care Excellence (NICE) guidelines (2011) developed following rigorous systematic review of the evidence base and expert opinion. NICE guidance is widely acknowledged as a 'gold standard' internationally and most teams in the UK practice according to these guidelines (National Institute for Health and Care Excellence, 2011). Services are largely based in either Child Development Services/Paediatrics (CDS), CAMHS, or tertiary services, including Tier 4/In Patient CAMHS.

In the absence of a confirmatory biomedical test, diagnosis requires assessment of the CYP in more than one setting, for example home, school and clinic. In line with most English language national guidelines evaluated in a recent systematic review (Penner et al., 2018a), NICE (2011) recommends this is best achieved using a multidisciplinary team (MDT) approach with representation from CAMHS. The core team should include a paediatrician and/or child and adolescent psychiatrist, speech and language therapist (SALT) and psychologist with the skills and competencies to determine whether a CYP meets diagnostic criteria (Wallace et al., 2013). Use of validated formal structured history and observational tools are recommended where needed but NICE recognises that no single tool is sufficient for diagnosis of autism (National Institute for Health and Care Excellence, 2013). Clinicians also need to consider differential diagnoses including other neurodevelopmental disorders, mental and behavioural disorders, regression of developmental skills, maltreatment and sensory impairment (National Institute for Health and Care Excellence, 2011).

Diagnostic assessment should enable clinicians to construct a profile for each CYP including intellectual ability, language and communication skills, motor skills, and mental and emotional health (National Institute for Health and Care Excellence, 2011) and identify essential areas of support and medical need for the individual and their family. However, diagnostic assessment can be time consuming and

costly; a recent study found assessments took a mean of 13 hours of professional time, costing £800 per patient (Galliver et al., 2017). Our recent study of 500 children's diagnostic journeys gave average figures of 15 hours, costing around £950 per child (Male et al., 2019). However, the current economic climate offers limited opportunity to increase financial resources to meet growing demand and is exacerbated by issues around recruitment and retention of suitably skilled staff. Additionally, current trends suggest future prevalence of autism may continue to rise resulting in even greater pressure on services and longer waiting times (Waugh, 2017, Baio et al., 2018).

1.3 The diagnostic pathway: capacity versus demand

Families frequently report negative experiences of the autism diagnostic pathway including long waits from referral to diagnosis, insufficient information, and lack of signposting and access to support post-diagnosis (Reed and Osborne, 2012, Crane et al., 2016). Journey times from referral to diagnosis vary significantly across the UK but a recent service review by the Care Quality Commission (CQC) and Ofsted (2017, p.10) commented on 'unacceptably long delays' waiting for diagnosis with families describing 'becoming even more frustrated and sometimes isolated, with little or no support.' There appears to be increasing pressure to deliver timely assessment set again increased demand for diagnostic assessment (Rutherford et al., 2018) but beyond the issues of demand and capacity, there are several factors relating to the child, their family or the professionals involved, which contribute to the time taken in reaching diagnosis.

The longest journey times reflect either complexity, adoption of a 'wait and see' approach, or family factors such as poor clinic attendance (Rutherford et al., 2016), or level of clinical expertise. Brett et al. (2016) identified the importance of case complexity in delaying age of diagnosis, with the presence of additional neurodevelopmental diagnoses such as ADHD (Kentrou et al., 2019) delaying diagnosis. At the same time, they found that increasing recognition of 'milder phenotypes' in school aged children, which are often more difficult to unravel diagnostically, often delays diagnosis whilst those presenting with language regression, or severe language impairment are generally identified at a much younger age (Brett et al., 2016). This study also identified family factors including socio-economic status and the presence of a pre-existing sibling with autism in the family which may reduce time to diagnosis.

With the impact of growing demand for diagnostic assessment, the resources required to deliver this, and the resulting lengthy journeys through this process and increased family stress (Reed and Osborne, 2012, Crane et al., 2016), a number of services have already adapted their pathways in an attempt to become more efficient. Approaches include single practitioner or abbreviated assessments (Penner et al., 2017, Rutherford et al., 2018, Whitehouse et al., 2018), both reducing the hours of

professional time involved (Penner et al., 2018b); the use of skill mix, for example replacing expensive doctor time with a nurse or allied health professionals (AHPs); training programmes to increase the skills of team members and referrers; organisational change and improving information gathering whether prior to accepting the referral (Rutherford et al., 2018) or even through the use of digital technologies (Jordan et al., 2017). Whether these strategies improve ability to deliver a timely and valid diagnosis, and whether there are unforeseen negative consequences is unknown and needs investigation.

1.4 Study rationale and the evidence gap

Timely diagnosis is important for the CYP, family and wider society (e.g. health, social care and educational professionals) for the following reasons:

- Diagnosis provides a clear explanation of an individual's strengths and difficulties (National Institute for Health and Care Excellence, 2011) and helps those involved in his/her care to better understand the needs of the individual and their family. This should enable parents to access appropriate health, social care and education services (Elder et al., 2017), support packages, and financial support such as Disability Living Allowance. In addition, CYP and families should receive timely access to interventions, whether early intervention programmes in preschool children, or appropriately adapted treatments in older children (National Institute for Health and Care Excellence, 2011).
- Autism diagnosis allows context for diagnosis of co-existing conditions, such as school refusal, attention deficit hyperactivity disorder (ADHD), anorexia and anxiety disorders (National Institute for Health and Care Excellence, 2011). Additionally, early ruling out of an autism diagnosis facilitates recognition of alternative explanations of the individual's difficulties.
- Early diagnosis helps to inform short, medium and long-term provision and planning of health and social care services, school and preschool provision both for the individual child, and more generally for the whole population (RCPCH, 2017).
- Early diagnosis and intervention has the potential to reduce costs, for example, one US study estimated early diagnosis and access to early interventions could deliver cost savings of $187,000-$203,000 per child for ages 3-22 years (Jacobson et al., 1998).

Currently, the evidence is not sufficient to identify: a) the most effective and efficient diagnostic pathways for CYP with possible autism which reduce the time taken from initial referral to completed diagnostic assessment, and b) how to deliver appropriate support to families throughout the diagnostic process. Clear and robust evidence is needed to identify which models have the greatest potential to meet the growing demand for diagnostic assessment in a timely, clinically valid, and family

friendly way. Given that some local providers have already reconfigured their services in an attempt to address these issues and deliver more efficient, and timely diagnostic services (Palmer et al., 2011, Male and Reddy, 2018, Male et al., 2020), this study will explore models of diagnostic service delivery that have been adopted across the UK, to determine which approaches function best for families and commissioners, in what contexts and identify factors, or mechanisms, that contribute to their success or failure. This should inform national policy, local commissioners, NHS trusts and local education, social care and third sector services.

1.5 Research questions

This Rapid Realist Review (RRR) is the first step in a national **R**ealist **E**valuation of **A**utism **S**ervi**Ce** **D**elivery (RE-ASCeD). The review aims to explore how particular approaches may deliver high quality and timely autism diagnostic services for CYP with possible autism; high quality is defined as compliant with NICE (2011) guidelines and timely is defined as a pathway lasting no more than one calendar year, based on previous work. Research questions for the RRR are as follows:

1. How do various models of autism diagnostic and support services address the differing needs of different service user groups and what contexts and mechanisms affect their ability to do so?
2. How do models of autism diagnostic and support services improve service user diagnostic experience?
3. What aspects of implementation, staffing and organisational context influence how models of autism diagnostic and support services operate?

2. METHODS

The complexity of autism diagnostic pathways in terms of differences in local service configurations and settings, lends itself to a realist review method that can tease out contextual factors, resources, and responses of those delivering and accessing the services. Realist reviews present evidence as programme theories (key features of the service with a description of what appears to be leading to certain outcomes) (Wong et al., 2013). These programme theories are supported by details of the context (C), mechanisms (resources and responses of those delivering and accessing the service) (M), and the intended (and unintended) outcomes (O). These are presented as CMO configurations. In using an RRR approach we worked backwards from the intended outcomes as identified by NICE guidelines (National Institute for Health and Care Excellence, 2011) and the NHS England Long-Term Plan (NHS England, 2019b).

What differentiates RRR from a full realist review is arguable but RRR is explicitly designed to engage those with expertise in order to accelerate the search process and validate findings (Saul et al., 2013). The wider RE-ASCeD project team have significant expertise in realist evaluation, paediatric neurodisability, child psychology and autism. The Expert Stakeholder Group consists of content experts, knowledge users and policy makers with a wide range of experience, including consultant paediatrician, speech and language therapy, child psychology, occupational therapy, third sector advocacy groups and patient and public involvement (PPI). However, stakeholder involvement does not replace the literature search but serves to supplement, tailor and expedite it (Saul et al., 2013, Willis et al., 2014).

Our focus is a clearly defined intervention (the diagnostic pathway), associated with specific outcomes (high quality and timely) within a particular set of parameters (Autism/CAMHS services in England); this is consistent with a realist focus on 'theory-driven, contextually relevant interventions that are likely to be associated with specific outcomes within a particular set of parameters'(Saul et al., 2013, p.3). Our initial Programme Theory, based on NICE (2011) guidance, the project team and Expert Stakeholder Group, states:

> *If there is a MDT assessment by a team with competencies in child neurodevelopment and mental health (context), then autism will be recognised as a complex condition that relies on detailed history and observation across settings (mechanism) to diagnose it. This will lead to accurate diagnosis, recognition of associated co-occurring conditions such as ADHD and intellectual disability (outcome), and the ruling out of complex differential diagnoses. This will also create, whilst not an explicit part of this project, an accurate picture of a child's strengths*

and needs to inform individualised packages of support and intervention through health, education and social care (outcome).

We followed the five stages of RRR (Saul et al., 2013), consistent with the RAMESES standards (Wong et al., 2013) for realist syntheses including developing and refining the scope of the review; searching and identifying information; extracting and appraising the evidence; synthesizing and interpreting the evidence; validation with expert stakeholders and dissemination. Table 1 provides a summary of realist terms. Figure 1 shows the iterative stages of this RRR, with information on key actions and involved stakeholders at each stage. Preliminary explanations will then be tested within later stages of the whole project.

Table 1 Realist terminology

Term	**Explanation** (Jagosh et al., 2011, Jagosh et al., 2014)
Context	Refers to the 'backdrop' of interventions, or anything outside the parameters of the intervention that might affect it, for example, shortages of community paediatricians. Context can also be understood as 'any condition that triggers and/or modifies the mechanism',[34].
Mechanism	The generative force that leads to an outcome of interest; usually hidden and context-sensitive. Mechanisms consist of intervention resources and how people respond to them.
Outcome	The outcome, intended or unintended, of a complex intervention such as timely assessment of possible autism in children. Outcomes can be initial, intermediate or final.
CMO configuration(s)	CMO configuring is a heuristic used to general causative explanations relating to outcomes. The process explores the relationship between an *outcome* of interest in a particular *context*; and the underlying *mechanism.* CMO configurations are a way of refining theory about aspects of the intervention, or the intervention overall.
Programme theory	This is the overarching theory of how a particular complex intervention may work; it draws on evidence, data and creative (retroductive) thinking to seek explanations of how, why and in what contexts, an intervention works. The initial programme theory is tested and refined in an iterative process throughout the RRR.
Programme areas	These are areas, or themes, within the overarching theory, for example, GP involvement in the autism referral pathway.
Middle range theory (MRT)	An explanatory theory that can be used to explain a complex intervention, or aspects of it. While CMO configurations are specific to the intervention under investigation, and underpin the programme theory, MRTs are more generic and have wider application.

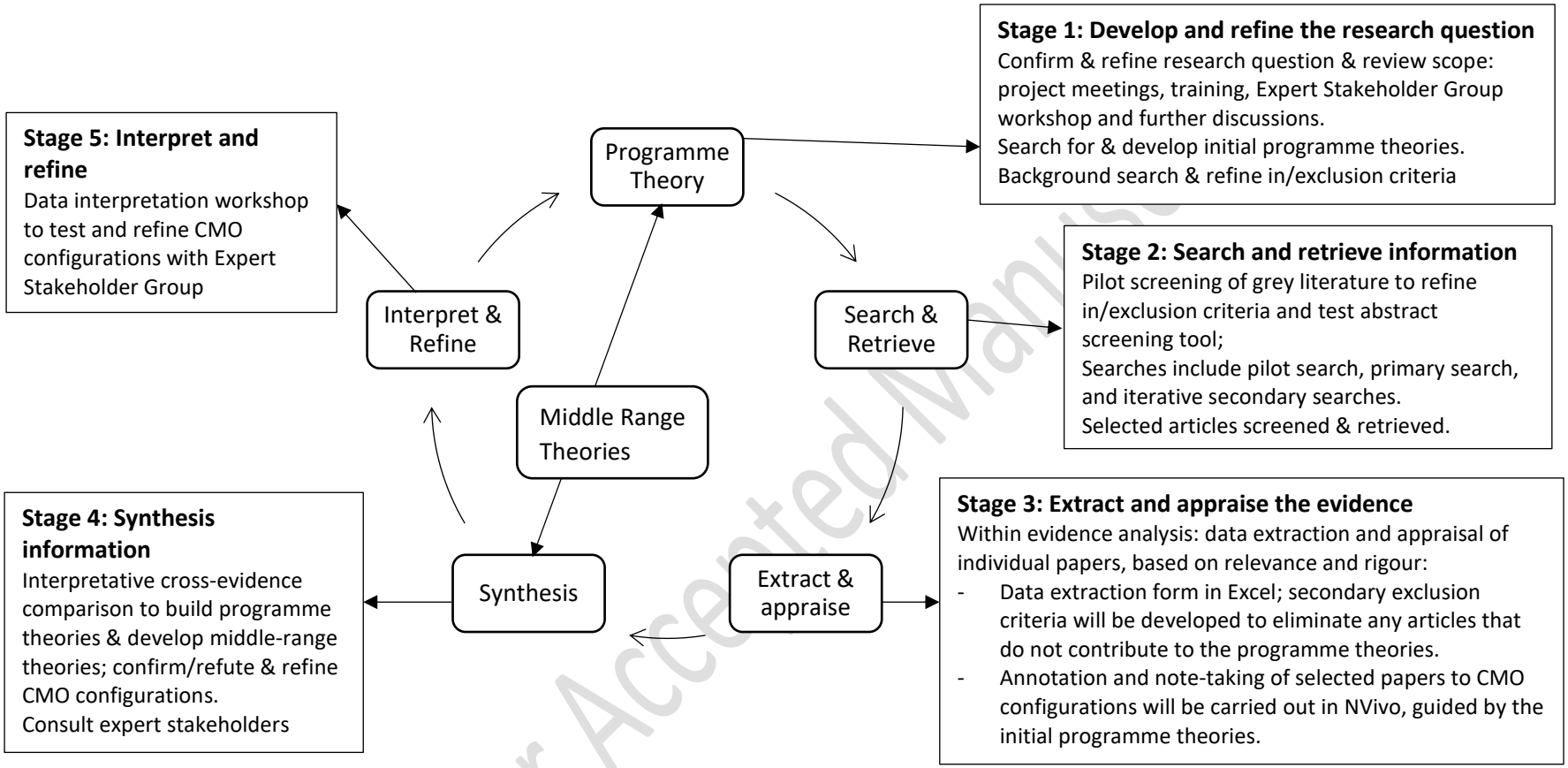

Figure 1 RE-ASCeD rapid realist review stages

2.1 Stage 1: developing and refining the research question (Oct-Dec 2019)

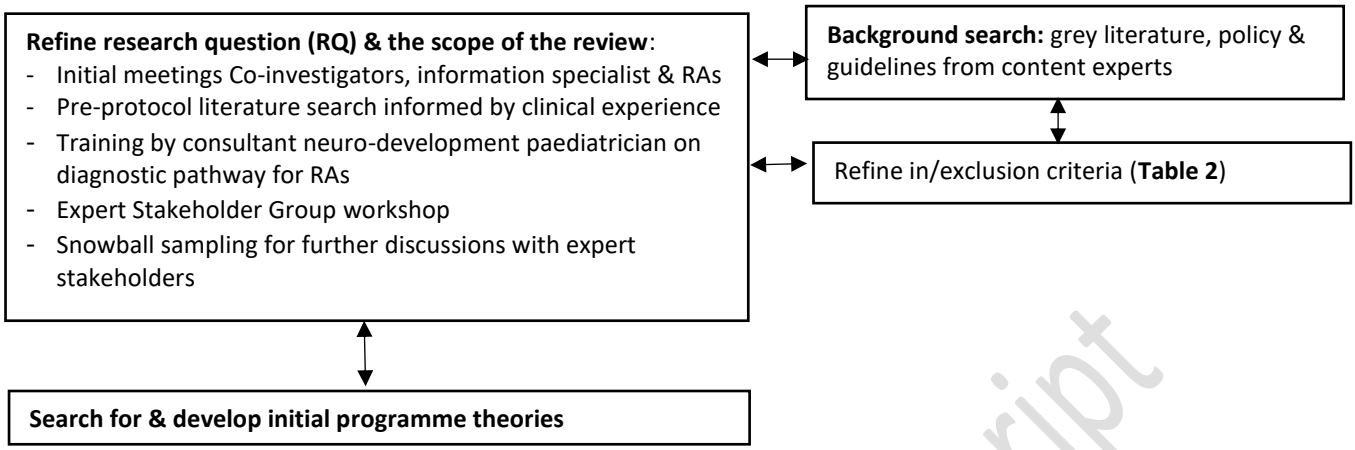

Figure 2 Developing and refining the research question

At the first stage, we confirmed and refined the research question and scope, and prioritised areas for investigation (Figure 2). This stage was built on discussions with our Chief Investigator (IM, Consultant community paediatrician), Co-investigators, NHSE and our Expert Stakeholder Group.

The RRR team (the core researchers working on the RRR within the larger project team) carried out preliminary work for Stage 1, as follows:

- An initial pre-protocol literature search informed by clinical experience (IM)
- Discussion within the RE-ASCeD project team to define the scope and remit of review
- Training on the diagnostic pathway for the research assistants (RAs) (VA, WZ) by a consultant neuro-developmental paediatrician, also a Co-investigator.
- Expert Stakeholder Group workshop to help confirm and refine the research questions, inclusion and exclusion criteria (Table 2), and identify salient documents (policy and grey literature) for review (Willis et al., 2014). Telephone discussions were carried out with expert stakeholders who were unable to attend the workshop.

The above Stage 1 activities function as a starting point to develop initial programme areas. Based on these preliminary work, the RRR team further discussed with the Expert Stakeholder Group before moving to stage 2. With support from our expert stakeholders, the background search included 129 grey literature and policy and guidelines papers.

Table 2 Inclusion/exclusion criteria

Primary inclusion criteria:
1. Children (preschool, primary or secondary school and adolescents) with Autism Spectrum Disorder OR Autism spectrum condition AND

2. UK healthcare system (England, Scotland, Wales and/or N. Ireland) AND
3. Published 2011 onwards AND
4. Relates to diagnostic pathway & model of service provision OR
5. Relates to assessment process e.g. single discipline (paediatric consultant) or inter-disciplinary

Primary exclusion criteria:

1. Non-UK based literature
2. Relates *only* to adult diagnostic pathway
3. Relates only to *tertiary* services
4. *Only* relates to treatment
5. Relates to support services only *after* diagnosis.

2.2 Stage 2: searching and retrieving information (Jan-Mar 2020)

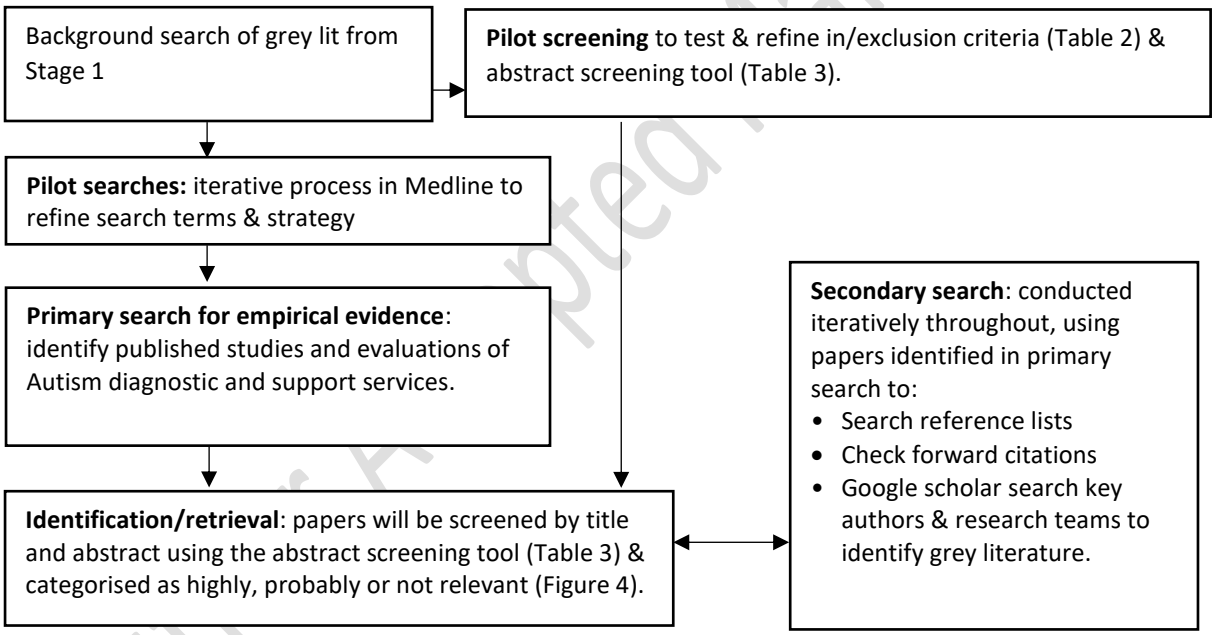

Figure 3 Searching and retrieving information

Stage 2 started with screening information from the background search carried out in Stage 1, which included national policy/guidelines and grey literature on local implementation of the diagnostic pathway (Figure 3). The search carried out by the information technologist were performed in three stages (Tsang et al., 2015):

1) **Iterative pilot searches** were carried out in Medline to refine the search strategy. Using the search strategies designed with support from Co-Is and experts, the pilot searches used a combination of text terms and synonyms appearing in the title, abstract and keywords in combination with index terms (MeSH subject headings) using the OR operator. These

concepts were then combined using the AND operator. By checking synonyms and further exploring MeSH index terms, the search was tailored to ensure relevant papers are retrieved. The strategy was modified to increase the sensitivity of the search by excluding some terms, including others, trialling MeSH terms, and dropping the keyword field and restricting to the abstract and title fields only. Limits are papers written in English and published after 2011, when the NICE guidelines for recognition, referral and diagnosis of autism in under 19s was published (National Institute for Health and Care Excellence, 2011). The search was restricted to UK only, given the specific NHS context, but the secondary search included papers from similar countries (USA, Canada, Australia, New Zealand) albeit with different healthcare systems to help elucidate our findings, and as suggested by our expert stakeholders.

2) **The primary search for empirical evidence** was used to identify published studies and evaluations of autism diagnostic and support services. The primary search used databases including Medline (Ovid), Embase (Ovid), PsycINFO (Ovid), Social Policy & Practice (Ovid), CINAHL Plus (EBSCO), Cochrane Library and Web of Science (Clarivate) with limits of dates (January 2011 to December 2019), language (English) and country (UK only). Each database varies with different index terms (or none) and searchable fields which means the strategy will be amended for each database. Appendix A shows detailed search strategies used for the primary search. Overall, we collected 211 articles (after duplicates removed) from the primary search in seven databases.
3) **Secondary searching was conducted iteratively** throughout the review with input from our Expert Stakeholder Group. Two reviewers used papers identified in the primary and background search to look through reference lists for relevant articles; check forward citations; and search key authors and research teams to identify further literature, using Google scholar. Prior to data extraction, we also consulted our expert stakeholders to ensure that we were not missing any key literature; we repeated this step at the consolidation workshop (Stage 5).

Figure 4 shows the steps and outputs of the background search of grey literature and primary search.

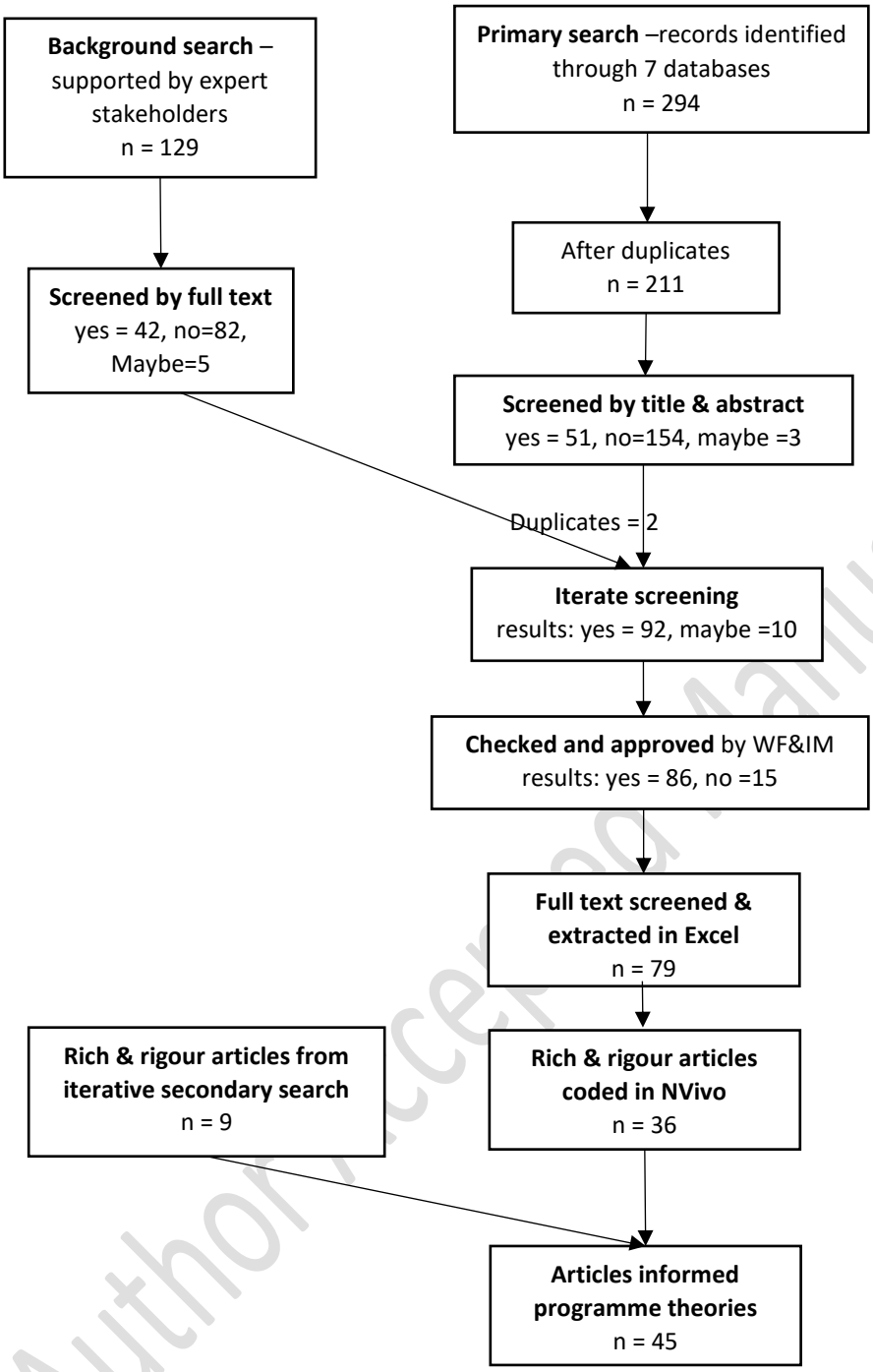

Figure 4 Flow chat of search & screening process of primary search, grey literature search, and iterative secondary searches

Two reviewers (VA, WZ) carried out an initial screen of primary search and grey literature search results by title and abstract using the abstract screening tool (Table 3) to identify articles as highly, probably or not relevant. This pilot screening helped further develop the abstract screening tool (Table 3) co-produced by the RRR team and expert stakeholders in Stage 1. We included studies of any design including randomised controlled trials, controlled studies, effectiveness studies, uncontrolled studies, interrupted time series studies, cost-effectiveness studies, process evaluations and qualitative studies.

To understand the complexities of the autism diagnostic pathway, we took account of literature reporting all stakeholder perspectives including those of children/adolescents, parents, clinicians, service managers and commissioners.

Table 3 Abstract screening tool

Ranking	Descriptions
Highly relevant	Primary focus is on diagnostic pathway whether the model is autism specific, Autism/CAMHS, integrated neuro-developmental pathway; AND
	Relates to certain *aspects* of diagnostic pathway e.g. skills mix OR
	Comments on implementation issues &/or contextual issues &/or outcomes OR
	Relates to quality or timeliness or cost-effectiveness of diagnostic pathway OR
	Relates to service user experience OR
	Relates to support services *up to* diagnosis OR
	Explores stakeholder perspective (commissioners, clinicians, other) *up to* diagnosis
Probably relevant	Some description of diagnostic pathway but not the main focus
	Little information on implementation, context or outcomes
	Limited reference to quality, timeliness or cost-effectiveness of diagnostic pathway
	Briefly refers to service user experience
	Briefly refers to support services up to (and/or post-) diagnosis
	Explores stakeholder perspective (commissioners, clinicians, other) up to (and/or post-) diagnosis
	Mostly not relevant but contains some 'nuggets'
Not relevant	Does not meet above criteria e.g. only focuses on post-diagnosis or adults

Due to the large number of inclusions and concerns about the consistency of screening between the two reviewers, all included literature was re-screened and further articles excluded using the secondary exclusion criteria (Table 4). Where no abstract was available, or reviewers were still unsure, full text was sourced, where available. A list of 92 'highly relevant' and 8 'probably relevant' documents were sent to the Chief investigator (IM) and one Co-Investigator (WF) who excluded 14 based on duplicates, date and relevance to the research question (Figure 5). The full text was retrieved

for all papers deemed relevant. Endnote was used to store and categorise the search results by the three relevance categories.

Table 4 Secondary exclusion criteria

1. Descriptive or irrelevant commentary on materials we already included; no added insights relevant to context or mechanisms
2. Specific tools in terms of assessment tools or psychometric properties – reliability/validity of the tool
3. Prevalence only studies
4. Studies only related to symptoms or aetiology
5. Articles about special needs in general, no mention of ASD (or ADHD)
6. Duplicate material of Co-Is' previous research – excluded by Co-Is
7. Only abstract is available and authors said 'Apologies- this was a conference paper, therefore I don't have a copy.'
8. The data collected or published on-line before 2011

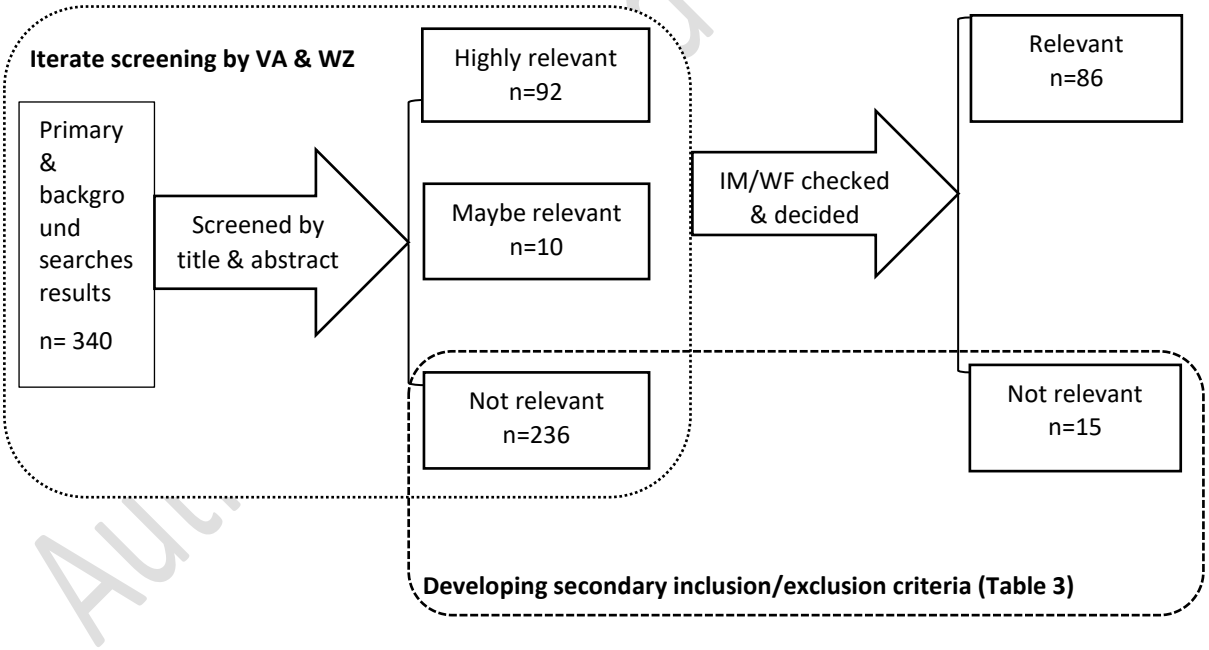

Figure 5 The screening process of primary search results

2.3 Stage 3: extracting and appraising the evidence (Mar-Apr 2020)

Within evidence analysis involved data extraction and appraisal of individual papers based on relevance, rigour and richness (Jagosh et al., 2011, Wong et al., 2013):

- Relevance refers to whether the evidence can contribute to theory building and/or testing; evidence can be highly relevant but not necessarily trustworthy.

- Rigour questions whether the methods used to generate the data are trustworthy and credible (or plausible); this includes 'nuggets of wisdom' (Pawson, 2006, p.127) in otherwise methodologically weak studies. Alongside quality, evidence was appraised in terms of how credible, or coherent, which involves examining the reasoning behind the argument and considering the coherence of the programme theory developed from the data (Wong, 2018).
- Richness refers to the existence and quality of causal insights and encompasses rich descriptions of context, process or outcomes (Jagosh et al., 2011).

Data extraction was carried out using a hybrid approach (Weetman et al., 2017). First, basic details from each included article (n = 79) were screened full text and recorded in a data extraction form in Excel; the tool includes information about the diagnostic pathway, service delivery characteristics, and any evidence related to intended (or unintended) outcomes (Tsang et al., 2015). Secondary exclusion criteria (Table 4) were developed to eliminate articles that do not contribute to the overall programme theory.

Second, highly relevant, rich and rigour articles were coded using the software package NVivo 12 (n = 36) to identify CMO configurations, guided by the initial programme theories. Included articles were imported into NVivo under 'sources'; the coding framework includes one node for each programme area (for example, assessments carried out with suitable skills mix expertise) and sub-nodes were created as appropriate. The reviewers coded relevant extracts of each paper, using annotation and note-taking to record comments related to context, outcomes and possible mechanisms (Weetman et al., 2017). Annotations and note-taking served as an audit trail and a record of our decision-making processes and rationales for developing our programme theories (Gilmore et al., 2019).

For both stages of data extraction and analysis, the reviewers (VA and WZ) reviewed three papers jointly and three independently and then compared findings; the remaining papers were split and reviewed independently by one reviewer. For twenty percent of papers, a series of calibration exercises were undertaken by the RRR Lead (PW). Also, when two reviewers were uncertain about the extraction or appraisal of a paper, this was discussed with the RRR Lead (PW).

Mapping the sources to test and develop the program theories, we divided papers involved in NVivo analysis into three categories: 1) key papers that described a model of service delivery (e.g. integrated neuro-developmental service) in detail and were conceptually rich, allowing us to refine our programme theories, 2) 'medium' papers that mentioned a model with some useful information but were not conceptually rich and of limited use in refining the programme theories, 3) papers with a few 'nuggets' (Pawson, 2006) relevant to the programme theories but otherwise of limited use. This helped us focus on key and medium papers that could contribute the most to developing a conceptual

framework (Pearson et al., 2015) and refining the programme theories. The reviewers also iteratively screened secondary search results and included nine relevant papers that contribute to our programme theories. Appendix B provides a transparent audit trail for the project team and expert stakeholders.

2.4 Stage 4: synthesising information (Apr 2020)

Our 'hybrid' approach to data extraction in Stage 3 aided the process of distilling context and outcomes thereby allowing identification of underlying mechanisms within each programme area. This helped make sense of our initial and evolving programme theory. Based on the analysis of individual paper, we then conduct cross-evidence comparisons to build programme theories and confirm/refute and refine CMO configurations (Table 5 and 6).

Evidence synthesis used interpretative cross-evidence comparison to interrogate the data: we moved iteratively between analysing specific examples (i.e. extracts from one study); comparing and contrasting findings from different studies; further searching to interrogate, confirm or refute CMO configurations; and refining the overall programme theory (MacDonald et al., 2016, Weetman et al., 2017). Jagosh (2020) refers to retroduction, or the activity of exploring observable patterns to discover underlying mechanisms. We involved the whole RRR team in this retroductive process to ensure validity and consistency of theory building (Wong et al., 2013).

We also consulted with expert stakeholders iteratively. Cross-evidence analysis allowed us to develop and refine CMO configurations, which we aligned with our research questions to build or search for existing explanatory middle range theories (MRT). At a practical level, we continued to use annotations and linked memos in Nvivo to maintain transparency and capture decision-making but also used simple tables in a Word document, one per programme area, to capture (multiple) CMO configurations which are linked back to the evidence, with commentary on possible MRTs (either existing MRTs and/or evolving ones).

2.5 Stage 5: interpreting and refining (Apr-May 2020)

We ran a data interpretation workshop with our Expert Stakeholder Group to test and refine CMO configurations, which further contributed to the refinement of our final programme theories and MRTs. At the workshop, the RRR team presented the review process and findings, with illustrations on how the evidence is 'falling' and building into the conceptual framework (Figure 6) and examples of key papers that contributed to the CMO configurations. Our expert stakeholders collectively revised the refined programme theories, provided feedback on each one with reference to their clinical and/or research experience and were invited to identify any missing parts in our programme theories. We also asked them to suggest any additional literature which helps elucidate the programme theories.

Based on the feedback collected from the data interpretation workshop, two reviewers checked and added new papers suggested by our expert stakeholders; refined the programme theories and conceptual framework; and wrote up the report together with the chief investigator (IM), Co-Investigator (WF) and the RRR lead (PW). Our expert stakeholders have been involved in commenting on our draft versions during the process.

2.6 Patient and public involvement

Engaging PPI representatives from the start, as part of our expert stakeholders Group, has enabled us to focus the review on questions that stakeholders are most interested in answering and enabled identification of salient documents for review, many of which are grey literature or unpublished (Willis et al., 2014). PPI involvement is integral to the overall project, has been embedded into the review protocol and was particularly helpful when synthesising and interpreting the data (Stage 5).

3: FINDINGS

This section provides the evidence used to inform each programme theory. Details of each study (36 papers from the primary and background searches and 9 from secondary search contributed into PTs) can be found in Appendix B. Programme theories are summarised in the conceptual framework (Figure 6), which also summarises the macro/meso context of autism diagnostic services. Programme theories 1-4 follow the referral pathway, from pre-assessment, to triage and referral management, the diagnostic assessment, management plan and follow up (Table 5). Programme theories 5-7 relate to issues that run through the whole pathway: working with families; inter-disciplinary and inter-agency working; and training, service development and evaluation (Table 6). Structural and organisational barriers impacting on the effectiveness of the autism pathway are wide ranging and include chronic underfunding, increasing caseloads, reduced training budgets and recruitment/retention issues, particularly paediatricians but also other disciplines with the appropriate expertise within the MDT (e.g. child psychiatrists, clinical psychologists, SALTS) (RCPCH, 2017, BACCH, 2019). Of these barriers, the increasing numbers of referrals was of particular concern to our Expert Stakeholder, given the expectation that services will meet demand to complete high quality assessments of an ever-increasing number of CYP. Other problems include lack of communication and co-ordination between services, predominantly health and education, inconsistent adherence to national guidelines and lack of leadership and clear direction (RCPCH, 2017, Rutherford et al., 2018, BACCH, 2019), all indicative of the need to have professionals with the appropriate training and expertise, as advised by our expert stakeholders. The subsequent difficulties encountered by parents/carers and children and young people (CYP) awaiting autism diagnosis are well documented, and include long waiting lists, being 'bounced' between services, limited follow up, and poor signposting to relevant services.

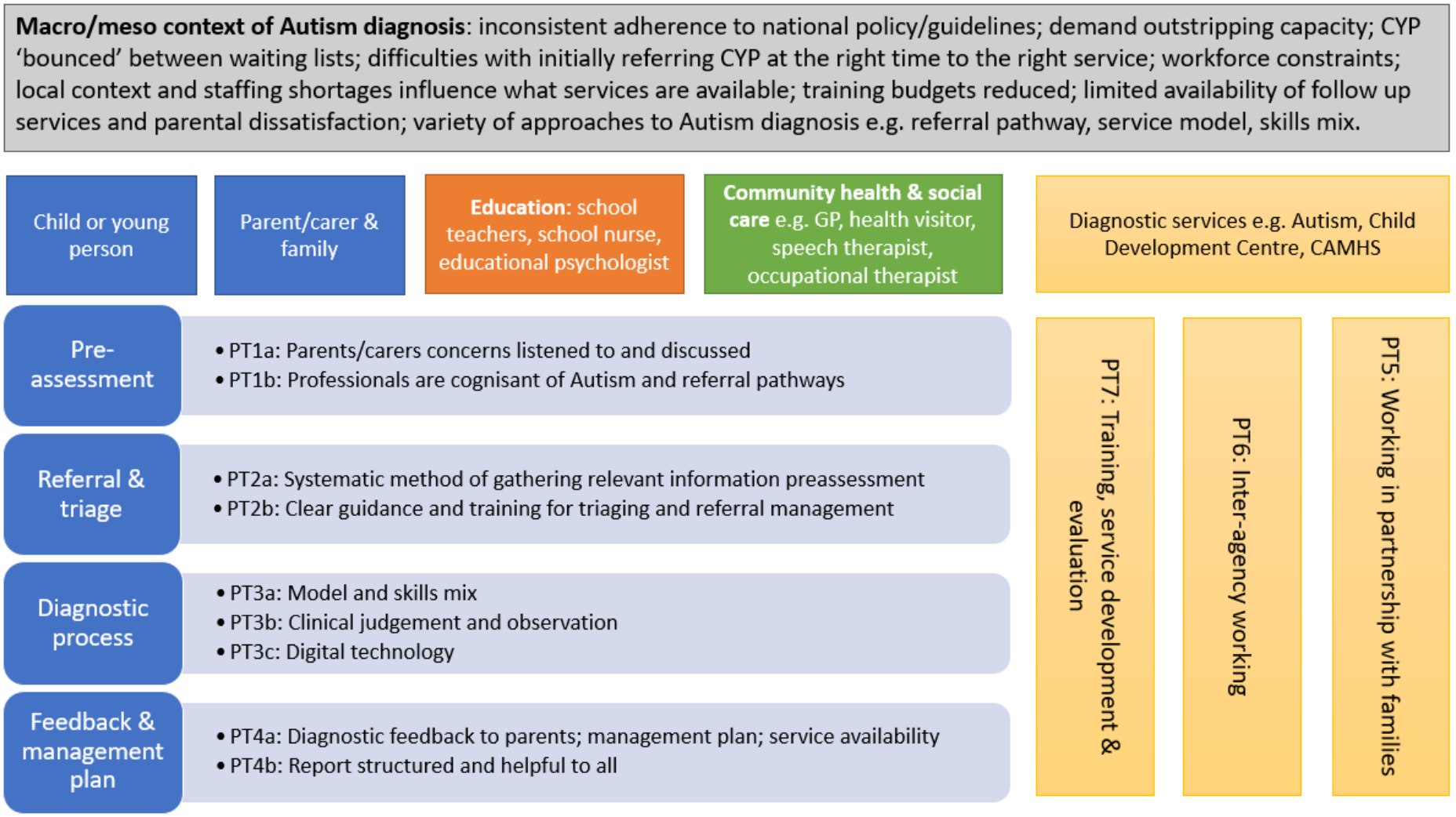

Figure 6 Conceptual framework – diagnostic assessments of CYP with possible autism

Table 5 Programme theories 1-4: stages along the diagnostic pathway

	Context-mechanism-outcome	Sources
Programme theory 1 - Recognition		
If frontline health and education professionals (e.g. GPs, teachers) are confident in recognising the signs and symptoms of autism, are cognisant of referral pathways and take parents/carers' concerns seriously then CYP will be referred to the appropriate service, in a timely manner, reducing parental frustration.		
1a. Parents/carers concerns are listened to and discussed	If frontline health and education professionals (e.g. GPs, teachers) take parents' concerns seriously (M), discuss and explain developmental behavioural concerns sensitively (M) and agree any actions to follow (M), then they will refer in a more timely manner (O) and parents will feel reassured with stress levels reduced (O). Also, if professionals at nurseries and schools (teacher or others) make a difference in "pushing" for a diagnosis or a specific form of support (M), then this will lead to timelier referral (O) and improve parental satisfaction (O) with the referral pathway. However, mis-diagnosis can be detrimental (C), so while parents should request referral for possible autism diagnosis (M) this has to be balanced against respecting professional expertise and enabling the development of a co-operative relationship (O).	NICE, 2011; Abbott et al., 2013; The Scottish Government, 2014; Rogers et al., 2016; O'Reilly et al., 2017; Unigwe et al., 2017; Crane et al., 2018; Dowden, 2018; Rutherford et al., 2018; Hurt et al., 2019.
1b. Frontline health and education professionals are cognisant of autism and referral pathways	If frontline health and education professionals (e.g., GPs, teachers) are trained in recognising the signs & symptoms of autism and referral routes (M), then their ability, confidence and skills in identifying children or young people (CYP) who need an autism diagnostic assessment will improve (O) and they will refer to the 'right' service in a timely manner (O). If proportionately fewer CYP go through the full process (M) then accessibility of services will increase (O), and the risk of unnecessarily raising parental concern over autism when it is not present will reduce (O). However, it is important to sensitively manage (M) a balance between supporting parents to accurately identify autism as early as possible, and not causing unnecessary concern amongst those who do not meet criteria for autism but may show some isolated Autistic-like features (O).	NICE, 2011; Reed and Osborne, 2012; Abbott, et al., 2013; The Scottish Government, 2014; Crane, et al., 2016; O'Reilly et al., 2017; RCPCH, 2017; Potter, 2017; Dowden, 2018; Hurt, et al., 2019; Ford, et al., 2019.
Programme theory 2 - Referral		

If autism diagnostic services provide clear guidelines for referrers on what information is needed and how to refer, then time will be saved at the triaging stage and fewer CYP who do not have autism will go through the full process.		
2a. Referral process	Referrals often lack relevant information; this adds to waiting lists and clinician time, as they gather appropriate additional information, delaying the diagnostic process (C). If referral is via a single point of access (for all neuro-developmental conditions and incorporating mental health expertise) (M) and referrers are provided with a systematic method of gathering relevant information from home and other settings preassessment (M) (e.g. proforma or digital assessment dashboard) and guidelines on how to do so (M), then referrers will know what information to gather, how to refer and what to expect following referral (O). When referrals are declined, the referrer should be provided with an explanation (M), advice for improving the referral (M) and/or other appropriate services to refer to. Collectively, these measures will contribute to reducing the waiting list and low diagnostic yield (low numbers of positive diagnoses) (O).	NICE, 2011; Carpenter 2012; The Scottish Government, 2014; MacKenzie, et al., 2015; Healthcare Improvement Scotland, 2016; Rutherford, et al., 2016; Rutherford, et al., 2018; Autistica, 2019; Tollerfield and Pearce, submitted.
2b. Triage	Services that triage referrals depend on having the necessary information (C). Cross-organisational triaging (e.g. monthly meetings with a representative from CAMHS, CCH and SLT), while time intensive, has several benefits including improved joint working (M response); a forum to discuss complex cases (M); improved compliance with the care pathway (O); only referrals with adequate information are accepted and therefore clinicians will use their time well (O); and this avoids referrals bouncing between agencies (O). Other approaches to triaging include an initial interview with an experienced clinician (M) who feels confident to identify CYP who clearly do, or do not, have autism; a community paediatrician carrying out a General Developmental Assessment/'Stage 1' Assessment, before referring to the MDT for further assessment, if needed (M). Although triaging and referral management requires very clear guidance and training for staff (M) it results in proportionately fewer CYP going through the full process who do not have autism (O) which reduces the risk of unnecessarily raising parental concern over autism when it is not present (O).	NICE, 2011; The Scottish Government, 2014; MacKenzie, et al., 2015; Healthcare Improvement Scotland, 2016; Rutherford, et al., 2016; Rutherford, et al., 2018; Hurt, et al., 2019; Tollerfield and Pearce, submitted.
Programme theory 3 - Diagnostic model		

There is wide variation in the model for autism diagnostic services and national staff shortages but these can be addressed with a structured and consistent approach, making best use of available staff and clinical expertise.		
3a. Model & skills mix	Current services have different condition-specific remits and models (e.g. Autism only, all neuro-developmental conditions, and/or integrated with CAMHS), catchment areas and commissioning agreements which raises challenges around capacity, care pathways and funding (C). Streamlining (M) the autism diagnostic pathway requires a structured and consistent approach (M) so that the number of assessments per individual are minimised, alongside developing efficient working and communication (e.g. shared proformas for report writing; on-line reports) (M), thereby saving resources (O) and reducing waiting lists. There is very little evidence to guide optimal service configuration (C) and the skills mix of diagnostic teams often relates to funding streams and the development of services over time (M). Core multi-disciplinary diagnostic teams are advisable (M) but there are national shortages of suitably trained professions including paediatricians and child psychiatrists who are the costliest members of the team (C). However, the role of professions that are available locally (e.g. SALT) can be extended by training them to carry out aspects of the assessment not requiring medical expertise (e.g. observational assessment) (M) which will reduce costs (O). Similarly, incorporating questions previously undertaken by psychiatrists into the parent interview (M) will free up time for psychiatrists to focus on complex diagnoses (O). Planning resources to meet need requires services to review their service configuration and skill mix (M) to accommodate demand within the available resources (O). Also recommended is ensuring that a core group of staff have dedicated autism time (M) and have shared skills for core aspects of autism assessment (M) to avoid overdependence on one clinician. However, disadvantages of MDT diagnostic assessment are that it takes longer and different professions may disagree (C). To reduce this added stress on families, professionals sometimes make their diagnosis independently (O). Some CYP referred for autism diagnosis may require mental health expertise and when unavailable, have to return to the waiting list for CAMHS (C). If the same Trust manages both community	NICE, 2014; Karim, et al., 2014; Gray, et al., 2015; Halpin, 2016; Healthcare Improvement Scotland, 2016; MacKenzie et al., 2016; Rogers, et al., 2016; Galliver, et al., 2017; Rutherford, et al., 2018. Ahlers et al., 2019; Autistica, 2019; Tollerfield and Pearce, submitted.

	paediatrics and mental health services (M), this potentially allows for a seamless transition, avoids duplicate waits and enables families to see all relevant professionals at the same time (O).	
3b. Clinical judgement	Diagnosis should involve interview, observation and recognised tools (C). Less experienced clinicians appear to prefer using formal extended tools compared to their more experienced counterparts (C). However, standardised tests lack subtlety and children may not meet cut-offs (e.g. atypical presentations) to receive a positive diagnosis. Clinicians often use their clinical judgement (M) to 'upgrade' the diagnosis so that the child is entitled to support (O). Many psychiatrists and paediatricians rely on the reports and observations of other professionals to inform their decisions while some, particularly educational psychologists, prefer to carry out their own observations within educational or home settings (C). This is valuable but time consuming; one solution (O) may be for professionals to only do observational assessment (M) if there are discrepancies between school and home reports. It is not always possible to provide a child with an accurate diagnosis at an early stage (C). Diagnostic uncertainty can lead to confusing and prolonged assessments (M) that may undermine both engagement and intervention (O). Therefore, reassessment after a specified timeframe (M) is necessary and the use of standardised assessments and observations (M) might be particularly helpful to aid diagnosis (O).	Carpenter, 2012; Karim, et al., 2014; Crane, et al., 2016; Rogers, et al., 2016; Rutherford, et al., 2016; Healthcare Improvement Scotland, 2016; Ford, et al., 2019.
3c. Digital technology	Children with autism sometimes feel an affinity for computing technology (C), as it is may be seen as a safe environment (M) to learn and practice skills that may be difficult in everyday life. The use of such technology in autism diagnosis is at an early stage (C) but shows potential, for example, using tablets/computers at school to collect observational data in a natural setting (M). If clinicians are able to access observations in advance (M), this would supplement other sources of data (O), save clinical time (O) and contribute to faster diagnosis (O). Telemedicine for autism screening &/or diagnosis is in the early stages of development (C) but shows some promise identifying individuals for further assessment (O) and early data suggest may be feasible and acceptable to parents and children (M).	Tryfona, et al., 2016; Jordan, et al., 2017; Juárez, et al., 2018.
Programme theory 4 - Diagnosis & support		

If parents understand the diagnostic process and feel supported this can moderate parental expectations. Feedback should take an assets-based approach and the management plan should be individualised, taking account of co-existing conditions. Reports should be timely and in a format that everyone finds helpful.		
4a. Diagnostic feedback to parents and CYP	Parents can find the diagnostic process stressful, and may fear the stigma attached to diagnosis, but anticipate that a positive diagnosis will act as a gateway to individualised information, advice, support, services and/or treatment (C). Receiving the diagnosis can affect parents' ability to absorb information but irrespective of the format (e.g. single professional or multi-disciplinary) (C) parents value: feedback that focuses on their child's strengths (asset based approach) (M) as this enables them to understand their child's needs (M), communicate these needs to others (O) and identify services to meet them (O); a structured and focused approach and the opportunity to ask questions (M); being put at their ease, listened to and given time to absorb information (M); and a positive and open parent-clinician relationship, established during the assessment process (M). Parental satisfaction is further enhanced (O) when the diagnosis results in an individualised management plan that identifies co-existing conditions (M); support post-diagnosis is co-ordinated and tailored to need (M); and appropriate services are available (M). Unintended consequences (O) include no autism or neurodevelopmental diagnosis which means parents may not be entitled to access condition specific services. Some CYP do not identify any benefits to diagnosis and fear being singled out as 'not normal' and subsequently stigmatised (O).	NICE 2011; NICE 2014, RASDN, 2011; Calzada, et al., 2012; Carpenter, 2012; Reed and Osborne 2012; Abbott, et al., 2013; Karim, et al., 2014; The Scottish Government, 2014; Halpin, 2016; Healthcare Improvement Scotland, 2016; Hennel, et al., 2016; Reed, et al., 2016; Rogers, et al., 2016; Crane, et al., 2018; The Scottish Government, 2018; Autistica, 2019; Hurt, et al., 2019;
4b. Report format	A standardised template for report writing, using consistent terminology, visual tools, enabled professionals to collate reports in a timelier manner and in a format that all found helpful.	MacKenzie, et al., 2016; Tollerfield & Pearce, submitted.

3.1 Programme theory 1 – Recognition

If frontline health and education professionals (e.g. GPs, teachers) are confident in recognising the signs and symptoms of autism, are cognisant of referral pathways and take parents/carers' concerns seriously then CYP will be referred to the appropriate service, in a timely manner, reducing parental frustration.

1a. Parents/carers concerns listened to and discussed

Parents describe difficulties being listened to by GPs, schools and nurseries (Crane et al., 2016). Several studies found that parents were often the first to notice atypical patterns of development or behaviour in their child and raise the possibility of an ASD diagnosis with clinicians (Abbott et al., 2013, Crane et al., 2016, O'Reilly et al., 2017, Potter, 2017, Dowden, 2018), often their GP, who either ratified or negated their concerns (O'Reilly et al., 2017). Many parents expressed frustration at the length of time it took their child to be diagnosed, and anger when professionals had previously discounted their concerns and/or given alternative explanations (Abbott et al., 2013).

Dowden (2018) explores the tension between some parents and professionals, creating barriers to diagnosis, either pre initial referral or in specialist services. Parents perceived professional reticence to diagnose as not believing them and only felt 'vindicated' (Abbott et al., 2013, p.375) when diagnosis was confirmed. While acknowledging parental frustration when a diagnosis is not given, professionals have to balance early referral and possible diagnosis against the consequences of mis-labelling (Dowden, 2018); one effective approach appeared to be managing parental expectations especially when access to condition specific services is based on diagnosis, rather than assessment of need (Hurt et al., 2019). NICE (2011) encapsulate these tensions and strike a balance between listening carefully to parents, appropriate referral, and importantly developing a co-operative relationship with parents. The guidelines recommend that clinicians 'always take parents' or carers' concerns… about behaviour or development seriously' (NICE, 2011, p.7) and when considering whether or not to refer, 'be critical about your professional competence and seek advice from a colleague if in doubt' (NICE, 2011, p.7) which links with onward referral. It may also help where support for the family is not diagnosis dependent and, as (NICE, 2014) suggests, through the development of a profile of the child's strengths, skills, and impairments including for example intellectual ability and adaptive behaviour, broader mental and neurodevelopmental health conditions, regardless of whether the child has autism, or not (NICE, 2014).

1b. Frontline health and education professionals are cognisant of ASD and referral pathways

The literature was consistent in supporting the need for greater autism awareness and training for frontline healthcare and educational professionals, particularly GPs and teachers, alongside training in how, when and who to refer (NICE, 2011, The Scottish Government, 2014, Crane et al., 2016,

Rutherford et al., 2016, O'Reilly et al., 2017, Unigwe et al., 2017, Dowden, 2018). However, professionals have to balance supporting parents to accurately identify autistic-like features as early as possible against causing unnecessary concern for those children who might display some isolated autistic-like features but are not deemed in need of assessment (Crane et al., 2016). Rutherford et al. (2016) recommended providing training for referrers that includes what would be expected (e.g. signs and symptoms, referral pathway) but also how to recognise autism in girls and differential diagnoses. While parents expressed frustration that teachers were not routinely taught about autism during or after their training (Hurt et al., 2019), we did not find literature to confirm or dispute this. GPs reported varying degrees of training in autism, pre- and post-qualification but still lacked confidence in identifying and managing those with a possible diagnosis (Unigwe et al., 2017). Confidence related to professional and personal experience, and to a lesser extent, training. Other factors explaining diagnostic reticence were lengthy delays between referral and diagnosis; lack of clarity surrounding referral and care pathways; ten-minute consultations being too short for such a complex condition; and lacking the resources to properly diagnose or look after CYP with autism (Unigwe et al., 2017). Opinions were divided about the remit of GPs in autism diagnosis from awareness of local services and signposting to taking on a specialist role, which most thought unrealistic given their skills and workload (Unigwe et al., 2017).

3.2 Programme theory 2 – Referral

If autism diagnostic services provide clear guidelines for referrers on what information is needed and how to refer, then time will be saved at the triaging stage and fewer CYP who do not have autism will go through the full process.

2a. Referral process

McKenzie et al. (2015) suggested that the most important predictor of assessment duration in CYP was the amount of information available in advance with more information reducing the number of contacts, the assessment duration and total time taken to reach diagnosis. The NICE guidelines (2011) endorse the need for information gathering from parents, schools and other agencies, and careful co-ordination to avoid duplication. A comprehensive referral also helps diagnostic services with managing referrals and triaging. Recommendations to improve the process included information leaflets for services including GPs, Health Visitors, nurseries and schools which describe the autism pathway, how to recognise autism, what information is needed, how to refer and what to expect following referral (Rutherford et al., 2018); using a systematic approach to gather information from referrers, for example using a proforma (Rutherford et al., 2016); designing a pack for education colleagues about contextual assessment, observation and reporting to inform the diagnostic assessment process (Rutherford et al., 2016); providing feedback when referrals are declined in order to improve the

quality of subsequent referrals (Rutherford et al., 2016); consultation with those involved in the child's care (NICE, 2011, Rutherford et al., 2018); and providing clear guidelines on what to include in a referral, such as history of developmental milestones, medical history and previous investigations (NICE, 2011). Additionally, Tollerfield and Pearce (submitted) recommend a digital assessment dashboard which can be pre-populated with, for example, parent questionnaires scored automatically, thus reducing time and ensuring all information is in one place.

2b. Triage

Not many papers discussed triaging in sufficient detail to elucidate underpinning mechanisms. However, some services involve a community paediatrician carrying out a General Developmental Assessment (GDA), or the equivalent in CAMHS of a 'Choice' appointment, before referring to the MDT for further assessment (Rutherford et al., 2018, Male et al., 2018, Autistica, 2019). Rutherford et al. (2018) described autism multi-disciplinary triage with staff from CAMHS or a community paediatrician and Speech and Language Therapy (SALT) assessment; cases are then allocated to an abbreviated (local) or complex (specialist) pathway (see 3a) which avoids duplication of waiting lists. Of those accepted onto the local pathway at triage and for whom assessment was completed, the majority were given a diagnosis of autism. Additionally, monthly triage meetings attended by a representative from CAMHS, Community Child Health and SALT, whilst time consuming, increased the amount of joint working across professional groups, provided a forum for discussion and ensured referrals were only accepted when there was sufficient information (Rutherford et al., 2018).

Carpenter (2012) suggested that an initial thirty-minute interview with an experienced clinician who is confident in their ability to distinguish between an individual who has autism and one who does not, and decide if the individual needs a detailed assessment, has significantly reduced the numbers of individuals needing full (specialist) diagnosis. While questionnaires for autism in CYP might be helpful, in that they prompted (less experienced) clinicians to consider the diagnosis, Carpenter (2012) cautions against relying on such screening tools as most, if not all, lack validity. Our expert stakeholders commented that carrying out an initial developmental assessment to cover broader issues than just autism in thirty-minutes would be highly challenging but were aware of others taking ninety-minutes or more.

Finally, the Newcomen Neuro-Developmental Service, Evelina London, is one example of a tertiary service that provides a single multi-disciplinary assessment completed in a half-day (Autistica, 2019). Every CYP receives the same assessment, with access to psychiatry if needed. While staff intensive, Evelina is able to gather comprehensive information from primary care, pre-assessment, to expedite the process. Other examples of similar tertiary services, suggested by our expert stakeholders, include Great Ormond Street Hospital, London (GOSH Foundation Trust, 2018) and the Complex

Neurodevelopmental Service, Newcastle (Cumbria Northumberland Tyne and Wear NHS Foundation Trust, 2018).

3.3 Programme theory 3 – diagnostic model

There is wide variation in the model for autism diagnostic services and national staff shortages but these can be addressed with a structured and consistent approach, making best use of available staff and clinical expertise.

3a. Model & skills mix

Very few primary research papers clearly delineated the service model (Karim et al., 2014, Gray et al., 2015, Rutherford et al., 2018, Hurt et al., 2019, Autistica, 2019, Male et al., 2020, Tollerfield and Pearce, submitted) and within these was wide variation in approach to diagnosis (e.g. referral pathway, skills mix and use of standardised tools) and the conditions included (e.g. autism/ADHD, autism/mental health). Autistica (2019) reported four multi-diagnostic pathways for neuro-developmental conditions but all had different remits. For example, Lambeth paediatric services, yet to be formally evaluated at the time of the report, rather than being diagnosis driven focused on profiling CYP's strengths and difficulties and then referred to other relevant services for formal diagnosis of possible conditions highlighted by the assessment.

Many services offer a single point of access but the conditions included varied. For example, Peterborough Integrated Neuro-developmental Services (Autistica, 2019, Male et al., 2020) referral is via the local Early Help Pathway and is usually completed by schools. From there, children of pre-school age see a community paediatrician for a GDA, before being seen by a multi-disciplinary team for further assessment, if needed, within the Child Development Centre (CDC). Older children and young people are accepted directly onto the pathway if there is sufficient evidence of a neurodevelopmental condition, following a referral (with a detailed description of the CYP and response to interventions that are already in place) through the Early Help Pathway. This approach, introduced in 2015, has significantly reduced waiting times and the referral-to-diagnosis rate has risen significantly (Autistica, 2019).

The same Peterborough service has integrated the pathway between CAMHS and community paediatrics which provides assessments for ADHD and autism, as well as supporting CYP with a previously diagnosed learning disability. The service provides mental health assessments for CYP with these diagnoses; if the individual has significant mental health needs, CAMHS can be accessed directly as the same NHS Foundation Trust manages community paediatrics and mental health services. The MDT includes consultant child psychiatrists, consultant community paediatricians, clinical

psychologists, nurses and support workers and access to SALT and occupational therapists thus combining a comprehensive skill mix with a single point of access (see 2a) and access to post-diagnosis interventions including a training programme for parents. This reduced the number of assessments per individual, saving time and money, and provided a better diagnostic experience for families (Autistica, 2019).

Tollerfield and Pearce (submitted) reported on their diagnostic service which was incorporated into CAMHS, with assessments completed by SALT and consultant child and adolescent psychiatry. To reduce the waiting list, additional funding allowed increased therapy time, but due to difficulties recruiting psychiatrists, this resulted in extended waiting times for psychiatry following assessment. In response, the SALT role was extended so that psychiatry time per case was reduced by approximately 70%, alongside significantly reducing the total clinical time required per assessment in the same time period (2013-18). Similarly, Rutherford et al. (2018) highlights the importance of planning resources to meet need which requires services to review their service configuration and skill mix to accommodate demand within the available resources. Also recommended is ensuring that a core group of staff have dedicated autism time and have shared skills for core aspects of autism assessment to avoid overdependence on one clinician (Rutherford et al., 2016).

While Tollerfield and Pearce (submitted) maximised the use of SALTs, McKenzie et al. (2016) study of adherence to the SIGN 98 Scottish guidelines found that only half of cases (40/80) received a comprehensive assessment of speech/language and communication skills carried out by a SALT with autism training; not all participating services had a SALT as part of their diagnostic team. One explanation was that if a CYP was verbal, a communication assessment might be regarded as unnecessary. Similarly, neither were all individuals considered for an assessment of intellectual, neuropsychological and adaptive functioning requiring access to clinical/educational psychology. The point being that all autism diagnosing services should 'have ready access to all professions who are needed to contribute to the ASD assessment and diagnostic process' (McKenzie et al., 2016, p.399), including clinical or educational psychologists, reflecting shortages in suitably trained professionals, particularly community paediatricians and child psychiatrists, impacting on waiting times for initial assessment and follow up (BACCH, 2019) and service development (RCPCH, 2017). Indeed, the NICE Quality Standard for autism diagnosis (NICE, 2014) requires that the 'Autism Team' should have a core membership of a paediatrician or child psychiatrist, clinical or educational psychologist and a speech therapist, with ready access to others, including neurology, occupational therapy and nurse support. This should enable the diagnostic team to understand the parents' concerns; to obtain a developmental history with focus on autism criteria; perform an assessment by interaction and observation of the child's social skills, sensory and repetitive behaviours; obtain a medical history and

physical examination; consideration of differential diagnosis; systematic assessment for conditions that may coexist with autism; and to develop a profile of child's strengths, skills, and impairments (NICE, 2014).

Several services take an assets-based approach which looks at CYP's skills as well as difficulties. Tollerfield and Pearce (submitted) developed a visual profiling tool that includes strengths, interests and skills. Each individual's unique profile of thinking patterns is considered within the context of other experiences and skills (e.g. early life experiences). Based on assessment information, a 'visual wheel' is produced, using a red, amber and green system, to display which aspects need consideration when developing management plans.

Rutherford et al. (2018) presented a clearly delineated multi-agency diagnostic pathway building on an early initiative (Rutherford et al., 2016). When the signs and symptoms of autism are easily identified during the GDA (stage 2 of the pathway) or at the point of triage (stage 3), CYP follow an 'abbreviated' pathway without onward referral to a specialist team. Using reports and observation made by local community clinicians, the diagnosis is confirmed by two or more clinicians, through mapping evidence from assessments to DSM-5 criteria. Other assessments are deemed 'complex' because of factors including co-occurring conditions such as ADHD, issues within the home, including exposure of the child to Early/Developmental Trauma, and family-professional disagreement. A case may also be perceived as complex when there is a mismatch between the skills, or skill mix, of a local team and the individual's presentation, for example when considering differential diagnoses. The parallel pathways result in fewer CYP who do not have autism going through the full process, reducing the risk of unnecessarily raising parental concern over autism when it is not present (Rutherford et al., 2018). However, abbreviated pathways could lead to issues of inequality, with families receiving different outputs depending on their route (Ahlers et al., 2019). Our expert stakeholders commented that this approach may also create issues over access for parents required to travel to the centralised 'complex cases' team, rather than a locally based service. Or equally, locally, that parents may question why some children are getting what has also unfortunately been called a fast track diagnosis.

Finally, disadvantages of MDT assessment are that it is labour intensive, costly, (e.g. Galliver et al. (2017) suggest 13 hours professional time costing £800/child), can take longer and can expose interprofessional disagreement, particularly between educational and medical paradigms. Karim et al. (2014) interviews with psychiatrists, paediatricians and educational psychologists found the different paradigms (educational versus medical) affected multi-professional and/or multi-agency working. For example, one paediatrician sometimes diagnosed independently and justified this on the grounds that involving all professions was too time consuming, adding to parental anxiety (Karim et al., 2014). However, where diagnosis was inter-professional, it relied on good relationships between individuals

which was not always the case. Halpin (2016, p.322)'s study of specialist nurses in community paediatric teams observed power differentials between members of the team; nurses did not challenge 'the diagnostic decision-making power of doctors and psychologists' while simultaneously expressing 'frustration' when their own professional role was not recognised as bringing a distinct clinical contribution. Despite these challenges, multidisciplinary practice is still strongly recommended as good practice within the NICE guidelines (2011), reflecting concerns over reliability and increased potential for individual bias for, or against diagnosis, with single practitioner assessment.

3b. Clinical judgement

An interesting theme within the literature considered the balance of clinical expertise against standardised assessments and which profession was best equipped (or could be trained) to carry out certain assessments. Less experienced clinicians appeared to prefer using extended standardised tools while those who routinely diagnosed autism were more confident and able to diagnose with less information (Carpenter, 2012). Karim et al. (2014, p.118) described how clinicians stated that they found diagnostic tools helpful, particularly in difficult to diagnose cases, but also found them 'cumbersome' and time consuming. Many clinicians reported not using any criteria to facilitate diagnosis, instead relying on clinical judgement. This appeared to relate to lack of objective or measurable markers for autism and tests that were deemed to lack subtlety or flexibility. This resonates with Crane et al. (2018), albeit written from an educational perspective, where those being assessed for autism compared the process with diagnosis for other conditions, where biomedical markers and a clear pathway appeared to provide reassurance. However, in some cases clinicians are simply not able to provide an accurate diagnosis at an early age and reassessment after a specified time frame is unavoidable (Crane et al., 2016). This diagnostic uncertainty and repeated assessments have the potential to undermine parental engagement and intervention and, in such cases, standardised assessments might be particularly helpful (Ford et al., 2019).

Rogers et al. (2016) refers to the literature around 'upgrading', whereby clinicians err on the side of a positive diagnosis when faced with uncertainty, in order to facilitate access to support, the rationale being that they were advocating for the 'best interests' of CYP and their families. However, upgrading most often happened in the *absence* of uncertainty, when clinicians were confident that the individual was on the spectrum but failed to meet cut-offs on standardised tools (e.g. atypical presentations and girls), reflecting the limitation of such tools. However, this raises concern around the accuracy and consistency of diagnosis but supports the case for service models that take a holistic approach to identifying strengths and weaknesses, rather than a yes/no approach to diagnosis (Autistica, 2019).

Karim et al. (2014) noted different approaches to the use of observation within and across professional groups but did not regard this as problematic, rather a reflection of clinical judgement. Educational

psychologists were likely to undertake their own observations in non-clinical settings, while many psychiatrists and paediatricians relied on the reports and observations of other professionals (e.g. occupational therapists) to inform their decisions. The timing of observations also varied, with some only being carried out if the information from standardised questionnaires was discrepant between different parties (e.g. parents and school) and was not in keeping with clinical observations (Rutherford et al., 2018).

3c. Digital technology

In contrast to the preceding section's focus on clinical judgement, Tryfona et al. (2016) focused on using mobile technology as an opportunity to collect data in advance that could contribute to assessment. The advantages were centred around CYP's familiarity with mobile devices; that those with autism may 'feel a strong affinity for computing technology, as it is seen as a safe environment to learn and practice skills' (Tryfona et al., 2016, p.252); that it would enable collection of data in a natural environment, such as home and school and allow for ongoing observations; and that this data, accessed in advance, could inform the appointment and save time. Observations could include different scenarios, such as family mealtimes or the behaviour that is of concern, however, the exact mechanisms of uploading the data on a shared and secure system need to be addressed.

Tryfona et al. (2016) also outlines various software options, such as sensors contained within mobile devices as an alternative to expensive eye-tracking software, specific educational games (as apps) with underlying algorithms that can identify the risk of autism through movement pattern analysis but all are in the early stages of development, lack implementation and are costly. At a more advanced stage, Jordan et al. (2017) have developed an automated story ('A Pirates Adventure') that scores emotional cognition in children. The aim is to use this alongside parental history and school questionnaire at initial clinical contact, to improve decision making about the need for full diagnostic assessment, potentially improving the patient journey and reducing costs. Our expert stakeholders also suggested that where the presence of ADHD is suspected, either as alternative diagnosis, or as comorbidity, the use of computer based Continuous Performance Tests such as the Qbtest (QbTech, 2020) may enable an objective measurement of attention, concentration, impulsivity and distractibility. The on-line results are instantly analysed and presented in a report that compares the individual's results with a group of people of the same age and gender who do not have ADHD, but the evidence to-date is limited.

Juárez et al. (2018) advocate their own TELE-ASD-PEDS tool designed by the Vanderbilt Kennedy Center (Nashville, USA) for use by providers with children under 36 months and carried out by families during a short telehealth assessment. A provider walks a parent through several basic tasks with their child which allows the provider to watch for the presence of autism symptoms. Administration is

meant to take 10-20 minutes. It covers the Autism Diagnostic Observation Schedule (ADOS) module 1 and some of module 2, and it is free, as are the resources and training. In terms of interaction with families, there is a conversation with parents about concerns and normal history taking. Using the tool to screen for autism, evidently saves time and identifies CYP who need further assessment. Of the forty-five children in this study, clinicians were confident with their diagnosis in all but six cases. Apart from minor challenges with technology, including low audio volume or video lag, the study suggested that it demonstrated feasibility, acceptability and accuracy of remote observation and early identification of autism with savings in time for providers and families. Similarly, our expert stakeholders report that the team who developed the ADOS have previously developed the Brief Observation of Social Communication Change (BOSCC) to monitor short term change for research purposes but are currently looking at how they can modify it to create a related video observation tool.

Since carrying out the RRR, Lord (2020) has provided guidance on how to adapt autism diagnostic assessment during social distancing (including ADOS) for remote use, demonstrating that the current covid-19 crisis has become a driver for telehealth approaches. There is already an existing telehealth publication with Module 4 (Schutte et al., 2015) with good psychometrics showing good agreement between face to face and remote administration. Similarly, our expert stakeholders report on trialling the TELE-ASD-PEDS tool. The main benefit appears to be that of appointing parents as co-professionals in the assessment alongside assessing the child in their own environment which provides rich data and can contribute to overall diagnostic assessment. Barriers include lack of appropriate IT equipment for families, in some cases addressed through loans from schools or charitable donations, and problems with loss of connection during consultations being commonplace. Parent reaction has been mixed, some like being in the safety of their own home while others have been reluctant to engage in video assessment. However, evaluation is needed to explore further sensitivity and specificity for best estimate clinical diagnosis and use in real practices with the full range of CYP referred.

3.4 Programme theory 4 – Diagnosis & support

If parents understand the diagnostic process and feel supported this can moderate parental expectations. Feedback should take an assets-based approach and the management plan should be individualised, taking account of co-existing conditions. Reports should be timely and in a format that everyone finds helpful.

4a. Diagnostic feedback to parents and CYP

The findings are mixed around the response to diagnosis with one paper suggesting that parents' reaction should also be assessed because parental stress can impact on the effectiveness of treatment (Reed and Osborne, 2012). It was not possible to make any judgement regarding single-professional or multi-disciplinary feedback beyond noting this varied (Gray et al., 2015). For example, Peterborough Integrated Neuro-developmental Services (Autistica, 2019) finalise diagnosis in an MDT meeting (joint CAMHS and community paediatrics) and one of the team then feedback to parents.

Several papers referred to parents' distress at hearing the diagnosis, even when anticipated (Abbott et al., 2013) and emphasised that care is needed to help parents through the diagnostic process, as parental stress levels may have repercussions on their own level of functioning which can impact on that of their child (Reed et al., 2016). One study, specific to fathers, found that the majority expressed a strong negative emotional response to their child's diagnosis (Potter, 2017) while another study, specific to mothers, suggested that while early diagnosis might lead to more rapid services and treatment, 'it may also leave mothers unable to develop coping mechanisms for living with this diagnosis' (Reed et al., 2016, p.7).

While some parents and young people were concerned about the stigma and/or discrimination that could result from diagnosis, many young people expressed 'indifference' about their diagnosis and did not attempt to find out about its meaning (Calzada et al., 2012, p.235). Parents regarded diagnosis as a gateway to services (Rogers et al., 2016) and 'ammunition' to push for support (Calzada et al., 2012, p.234). Diagnosis also helped them understand their child and brought relief that they were not to blame (Halpin, 2016).

Parents find it hard to absorb information during the feedback session, partly due to the emotional impact but also due to 'information overload' (Abbott et al., 2013, Hennel et al., 2016). They value practical approaches to help them absorb information such as a structured approach to feedback; opportunity to ask questions; being offered a further appointment for more discussion; and clinicians recognising their child's strengths, as well as difficulties, relating to an assets-based approach (see 3a). The intuitive approach of clinicians was also important – giving parents time to absorb information, listening to them, responding sensitively and having transparent (reciprocal) dialogue (Abbott et al., 2013). Hennel et al. (2016) provides further practical suggestions, including encouraging parents to bring a support person; providing written information to aid their understanding and help explain the diagnosis to others; and encouraging families to explore evidence-based websites, with the caveat that expectations around service availability need to be managed (Hennel et al., 2016).

Provision of information 'should always be viewed as a two-way process' (Healthcare Improvement Scotland, 2016, p.42) and whether verbal or written, professionals need to use consistent and

straightforward terminology when talking to parents (Karim et al., 2014, Healthcare Improvement Scotland, 2016), especially when the diagnosis is uncertain (or borderline according to current diagnostic criteria) and information must include short- and long-term implications, tailored to the individual's age, ability level and cultural background (Healthcare Improvement Scotland, 2016, Crane et al., 2018). Northern Irish guidelines (RASDN, 2011) recommend that as well as a written copy of the assessment and diagnosis outcome, a tailored intervention care plan is co-developed and outlines key contacts; which treatments/interventions will be offered and why; what support will be available; information about relevant agencies; and the contact details of a 'link person'. Similarly, NICE (2011, p.15) recommends documenting the individual's 'strengths, skills, impairments and needs that can be used to create a needs-based management plan, taking into account family and educational context', also alluding to an assets-based approach. Similarly, NICE (2014, p.10) also endorse a 'personalised plan' that is developed and implemented in partnership family and carers and the MDT needs to work with the family to implement the personalised plan alongside any other interventions or care the family are receiving (NICE, 2014).

4b. Report format

Although Tollerfield and Pearce (submitted) was the only paper to give detailed information on the report format, as distinct from general feedback, our expert stakeholder group advised that this was a distinct facet of this programme theory. The service used a report-writing tool producing a semi-automated draft report combining quantitative and qualitative information taken from a digital assessment dashboard (see 2a) which enabled the team to improve the quality and speed of reports. These were available for parents within a few days, enabling them to check that the content accurately reflected information given during the assessment, improving partnership working. The visual profiling tool created immediately after assessment also provides a concise visual aide for understanding, explaining and communicating the profile of each individual, including strengths.

Table 6 Programme theories 5-8: relevant to all stages of the diagnostic pathway

Programme theory 5 - Working in partnership with families Parents find the diagnostic pathway stressful so find it helpful to have a single point of contact; to be provided with explanations about the process; and to be included in decision-making.		
5a. Parent/carer as co-experts in the diagnostic process	Contributing to the patient-professional tension is a debate around who is the expert (C). Parents expect to be listened to during the diagnostic process and their concerns taken seriously because they 'know' their child (C); if they feel belittled and/or do not understand the process or terminology (Ms), they will disengage from the process (M) and/or resist alternative diagnosis (O) which will have a detrimental impact on the parent-professional relationship (O). Professionals need to explain the diagnostic pathway and acknowledge that it is enhanced (O) when expertise is integrated with the perspectives of the individual and their family (M). Parents want to have a transparent and honest dialogue with professionals (M) and be involved in key decision-making (O).	Gregory, et al., 2013b; Rogers 2016; Healthcare Improvement Scotland, 2016; Crane, et al., 2018.
5b. Supporting parents/carers	Some parents perceive the system as poorly co-ordinated and feel it necessary to take charge of organising diagnostic and support processes. However, a consistent point of contact within the system would provide emotional support and enable parents to be kept up-to-date (O). When professionals explain the diagnostic process in advance and how long it will take (M), this improves parental satisfaction and can moderate expectations (O). Non-attendance at appointments is frequent (C) and services need to have systems in place to reduce it, for example using reminders, opt-in systems and a support contact to facilitate attendance (M). By increasing attendance levels, this will reduce service costs and waiting times (O). When contact with professionals during diagnosis has been perceived by parents as unsatisfactory, this may lead to subsequent treatments undertaken by the child being less effective than they otherwise might have been (C). Satisfaction can be improved by managing the process in a thoughtful and sensitive manner (M); clearly explaining the diagnosis (M); and demonstrating a high degree of knowledge and empathy (M). Also, if some professionals (e.g. nurses) provide advocacy for parents' views during assessment (M) and well-organised parent/carer groups are established (M), parents' concerns are more likely to be heard and parents will be empowered to speak up for themselves (O).	Calzada, et al., 2012; Abbott, et al., 2013; Gregory, et al., 2013b; NICE 2014.
Programme theory 6 - Inter-agency working		

If "experts" including people with autism, carers, professionals and specialist organisations work in partnership and the knowledge generated is effectively embedded into local services, this will build capacity, improve parent/CYP satisfaction and support planning of services both locally and nationally.		
6a. Macro-Meso level	A multi-disciplinary, inter-agency and holistic approach is essential (M) given the subjective nature of diagnosis and the significant differences in presentation of CYP with autism (C). However, there are multiple barriers to inter-agency working at all levels, particularly a hierarchical relationship between education and health (C), with education practitioners delivering daily interventions but having to rely on healthcare professionals to issue diagnoses to release additional funding or support. Macro-level approaches to ameliorate these barriers include: setting up a national 'whole life' autism strategy that co-ordinates multi-agency planning (M); a national approach to support school pupils with autism (M); clear standards of training and expertise (M) for all service providers offering services for those with autism, and access to specialist training; positioning (strategically and/or physically) autism services alongside other CYP's services (M), as this enables the development of a shared understanding which promotes effective joint-working (O) and is particularly useful where CYP are at risk; a more integrated care pathway with additional ringfenced funding (M). If teams are supported to structure and deliver services in a flexible, creative, 'can do' approach at all levels from the clinician working on a day-to-day basis, to cross agency working, up through middle and senior management (M), then the experience of parents, children, clinicians and referrers would be improved (O). If partnership working across organisations develops and consolidates a combined skill set (M), has mechanisms in place to share information (M) and holds regular networking and multi-agency professional meetings (M), then this will support the development of a shared understanding of CYP, their support needs and those of their parents (e.g. negotiating with the wider system) (O).	NICE, 2011; Gregory, et al., 2013a; Karim, et al., 2014; NICE, 2014; The Scottish Government, 2014; Gray, et al., 2015; Healthcare Improvement Scotland, 2016; Rogers, et al., 2016; Galliver, et al., 2017; Hayes, et al., 2018; The Scottish Government, 2018; Williams et al., 2018; Hurt, et al., 2019; Tollerfield and Pearce, submitted.
6b. Micro level	Multi-agency working (M) is designed to minimise variations and enhance the engagement of all services (C). Improved co-ordination between health, education and local authorities (M), at the level of individual diagnostic assessment would help reduce the time taken from referral to diagnosis, improve parental perceptions of support following diagnosis (O) and, with clear documentation (M), improve information flow between involved parties (O). Opportunities to enhance multi-agency working include a "one stop shop" coordinator for children with ASD (M) and split posts for staff which can act as bridges between different parts of the system or	NICE, 2011; Calzada, et al., 2012; Gregory, et al., 2013b; The Scottish Government, 2014; Tollerfield and Pearce, submitted.

	different organisations (M), aiding understanding and communication (O). One opportunity to build links with relevant (voluntary) organisations (O) is to rent space, such as a community clinic, to carry out ASD assessments (M) but it needs to be an environment suited to the needs of children with ASD. However, when CDTs are based in a dedicated CDC (M), they are more likely to have implemented good practice recommendations including recommended team working and family communication standards (O). If ASD diagnostic services establish clear pathways, including detailed data on the use of time and tools at each stage of the process (M), this will improve effectiveness in assessing, diagnosing and supporting children with autism (O).	
Programme theory 7 – Training, service development and evaluation Based on their needs, skills and knowledge for autism diagnostic assessments and working with families, health and community professionals should have access to tailored training, service development and service evaluation.		
7a. Training for professionals working with CYP in community settings	Training in many organisations is "ad hoc", varies widely and may have low priority given financial constraints (C); multi-agency training is limited (C). Clinicians working with CYP with developmental delay, speech, language and communication impairments and mental health difficulties will come into regular contact with children with autism, as will frontline staff in generic children's services (e.g. nurseries) (C). If multi-agency training for professionals is provided (M), with a targeted and coordinated approach across organisations (M), a wide breadth of coverage of basic training can be achieved (M) and awareness and training geared to the needs of managers as well as front-line staff (M). This will increase the local skill set of people who regularly work with children who may have autism (O). Another approach is to develop a detailed framework, mapping staff skills and knowledge for autism diagnostic assessment at different levels (informed, skilled, enhanced and expert practice levels) (M). The levels of skill required by different staff depend on the nature, extent and likely impact of daily contact with individuals with autism (M), rather than defining levels specific to profession or position in a service. The framework can be used by individuals, organisations or training providers to identify current or future training needs at different levels (O).	NICE, 2011; Gregory, et al., 2013a; NHS Education for Scotland, 2014; The Scottish Government, 2014; Rutherford et al., 2016; Rutherford et al., 2018; The Scottish Government, 2018.
7b. Training for health professionals working in autism services	Training budgets have been reduced (C). If professionals working in autism services are provided with crucial supports, including backing for training, funding for a specialist library and practical resources (M) as well as access to supervision, links with other experienced professionals, and an open team culture of sharing ideas (M), then they will be able to work with CYP in the most skilled and effective way (O). As	Gregory, et al., 2013a; Autism ACHIEVE Alliance, 2014; Rutherford et al.,

	above, training programmes need to be tailored to the level of competencies required (i.e. enhanced and expert practice levels) (M). Training activities could include observing in a (tertiary) autism clinic (M) to develop skills and confidence (O); 'buddy up' with more experienced staff (M); regular Continuing Professional Development sessions for the team to review training needs (M); developing an explicit plan for succession planning and training needs (M); and a national forum to share experiences and knowledge, including people with autism and their families (M). As more staff become better trained in, for example, the use of standardised autism assessment tools (O), there will be a higher degree of consistency between local and specialist teams (O).	2016; Rutherford et al., 2018.
7c. Service development & evaluation	Structural and organisational barriers impact on the effectiveness of the autism pathway (C) and as services have become increasingly overburdened, clinicians have little time to engage with service evaluation and development (C). If services plan resources to meet need, based on audit data, for example reviewing service configuration and skill mix to accommodate demand (M) and make efficient use of administrative support to free up the diagnostic team (M), then time allocation and quality of autism services will be protected within resources and available capacity (O). Services should maintain or develop efficient systems of collecting information about referrals, waiting times and outcomes, for example using a guidelines checklist at the front of each patient file (M); data can be collated (M) for senior managers and commissioners to evidence shortcomings in staffing and resources (O). Suggestions to help promote service development and embed changes into practice (O) include having one person to lead/champion change (M); generating research within clinical teams (M); encouraging practitioners to co-create contextually sensitive solutions (M) in a cyclical process of service evaluation and development; and drawing on 'experts' within the field, including people with autism, carers and specialist organisations who could support local service development if identified and connected into the process (M).	The Scottish Government, 2014; Rutherford, et al., 2016; RCPCH, 2017.

3.5 Programme theory 5 – Working in partnership with families

Parents find the diagnostic pathway stressful so find it helpful to have a single point of contact; to be provided with explanations about the process; and to be included in decision-making.

5a. Parent/carer as co-experts in the diagnostic process

There is some overlap with PT4a, feedback to parents at the point of diagnosis, but our expert stakeholders advised that this was a distinct element of this programme theory because it highlights the importance of partnership working with families throughout the whole diagnostic pathway and beyond. To ensure the quality of the process, 'expertise from several perspectives needs to be integrated: that of the individual, their family, and the professionals' (Crane et al., 2018, p.3762) which acknowledges, as elsewhere (Gregory et al., 2013b, Rogers et al., 2016) that it is important to understand parents' views, as the experts on their children, but that professionals are the experts on autism. However, our expert stakeholders regarded this as contentious, because some parents have considerable expertise in autism, for example, having another child already diagnosed with the condition, and some clinicians are inexperienced. What matters is that professionals promote open, honest dialogue, so that the expertise of both parties is integrated, parents are involved in decision-making, and feel valued as 'co-experts' in their child's care (Rogers et al., 2016). This helps engage families and when differences of opinion arise, allows negotiation without fear of the relationship disintegrating (Gregory et al., 2013b).

5b. Supporting parents/carers

Exploring the key predictors of parental satisfaction with the overall diagnostic process, the stress of the process, including the wait, was found to play a key role in overall dissatisfaction (The Scottish Government, 2014, Crane et al., 2016, The Scottish Government, 2018, Hurt et al., 2019). When parents understood the diagnostic process in advance, including that the length of time taken was 'due to thoroughness and the need for several different professionals to make assessments' this improved their satisfaction and acted to moderate expectations (Abbott et al., 2013, p.373); one clinician had pre-warned parents about how long the assessment would take and had given them a timetable for the assessment process which parents found particularly helpful.

Parents offered support following diagnosis were, unsurprisingly, more satisfied than those who were not. Many parents reported a lack of support (practical and emotional) or were dissatisfied with what was provided; some stated that support was only forthcoming when they reached a crisis (Crane et al., 2018) or that help was withdrawn due to financial constraints (Crane et al., 2016). A simple (and cost-effective) suggestion to improve satisfaction is to tailor links to relevant services, for example arranging a follow-up appointment with occupational therapy, rather than simply signposting parents

towards generic services or omitting to explore the range of services that might prove useful (Crane et al., 2016).

Gregory et al. (2013b)'s paper relates to a CAMHS team specifically for children with disabilities, including autism, that supports parents beyond diagnosis. However, the lessons are pertinent - by co-creating a relationship with families, clinicians can assist them to 'strengthen' and develop strategies to manage the multiple difficulties they are likely to encounter (Gregory et al., 2013b, p.74). As part of acknowledging the importance of the relationship and the stressful situations families are managing, the service has no standardised rule for non-attendance. Instead, clinicians review parents' wish for input rather than equating non-attendance with non-engagement. The service also meets families wherever most convenient, including home, school or other community locations so that problems can be observed in the relevant context. This also reduces non-attendance at clinic appointments and circumvents transport issues (Gregory et al., 2013b).

Reducing non-attendance can decrease service costs and waiting times. Factors affecting non-attendance include forgetfulness, fear, transport difficulties, inconvenience of time/place, long waiting times and administrative errors (Rutherford et al., 2016). Suggested solutions with some evidence of success include providing information pre-appointment to engage parents; using patient driven appointment scheduling; having systems to pre-empt non-attendance (opt in letters, phone calls); reducing the number of inappropriate referrals, so that those offered appointments are those most likely to benefit; providing the service as locally as possible; and enlisting a support contact to facilitate attendance (Rutherford et al., 2016).

The involvement of many different professional systems across health, social care and education can be confusing and parents may need support with negotiating the system although some parents report taking on the role of co-ordinating diagnostic and support processes. Either way, having a named contact person, 'case coordinator' (NICE 2011) or 'keyworker' (NICE 2014), to negotiate and co-ordinate can reduce stress, increase engagement and enable parents to identify the most helpful person to work with for any particular difficulty (Gregory et al., 2013b). One option is for the school, in conjunction with the paediatrician, to be a central point of contact for parents and other services throughout the process (Autistica, 2019). However, our expert stakeholders commented on the lack of literature appertaining to the needs of parents with autism or a learning disability, or with other children already diagnosed with autism, and that support should be tailored to their specific needs.

3.6 Programme theory 6 – Inter-agency working

If "experts" including people with autism, carers, professionals and specialist organisations work in partnership and the knowledge generated is effectively embedded into local services, this will build capacity, improve parent/CYP satisfaction and support planning of services both locally and nationally.

6a. Macro-Meso level

Commissioning of separate autism and ADHD pathways, the former with Child Development Teams (CDTs) and the latter with CAMHS, results in children having to go through both pathways despite considerable overlap of assessment (Male et al., 2020). Integrating the pathways into a single assessment process potentially saves time, costs less and the time taken to reach diagnosis may be reduced, depending on variables such as case complexity. However, CAMHS and CDTs often sit in different healthcare trusts, each commissioned to deliver specific pathways, and failing to factor in the frequency of co-occurring conditions (Male et al., 2020). For about two-thirds of local authorities in England, co-working with CCGs is helped by having a simple relationship of one local authority to one CCG; however, over one-third of local authorities relate to more than one CCG and a small number work with four or more CCGs (Dowden, 2018). Similarly, co-working is helped by having a single NHS provider trust for CAMHS and CDC services (Male et al., 2020). Despite the drive for integration, Williams et al. (2018, p.367) assert that there has been little progress in integration over the past 20 years, citing their own Northampton CDC service as an example, where the social worker, occupational therapist, and educational psychologist have been withdrawn and no longer contribute to the assessment process. However, Galliver et al. (2017, p.4)'s cost analysis of autism diagnosis found that the length of time spent by doctors in diagnostic assessment appeared to directly influence (increased) cost compared to other disciplines, reflecting their hourly rate, thus 'allowing allied health professionals to carry out parts of the assessment not requiring doctor's skills…could save costs' and contribute to addressing the shortfall in community paediatrians (BACCH, 2019).

Most guidelines refer to MDTs as best practice for autism diagnostic assessment and follow-up but lack recommendations about how roles within (or across) teams are negotiated and how disagreement is resolved (Hayes et al., 2018). This resonates with interprofessional disagreement, mentioned earlier (PT3a), but we found no evidence of how to circumvent such problems at the macro/meso level. Where teams did work well together, it appeared to be based on good personal relationships at the micro-level (Gregory et al., 2013a) and a positive attitude, whereby teams adopt a flexible, creative, 'can do' approach at all levels from frontline staff working with families, across networks and with the support of management (Gregory et al., 2013a, p.70).

However, the above refers to healthcare settings and Hurt et al. (2019, p.218) demonstrated how 'health and educational systems operate independently with little crossover of activity', with parents taking on the role of coordinating services to try and link the two independent pathways. The expectations of the different professional groups were unclear and it was not apparent they understood their specific roles within the pathway. There also appeared to be a hierarchical relationship between education and health, with the former delivering interventions on a daily basis but 'having to rely on healthcare professionals to issue diagnoses to release additional funding or support'(Hurt et al., 2019, p.218).

NICE (2011, p.5) recommends that a 'local autism multi-agency strategy group should be set up, with managerial, commissioner and clinical representation from child health and mental health services, education, social care, parent and carer service users, and the voluntary sector' with a lead professional responsible for the 'recognition, referral and diagnosis of children and young people'. Furthermore, commissioners 'from across health, social care and education agencies' should work together to enable best practice, including implementing personalised plans (NICE, 2014, p.22). More recently, Male et al. (2020) suggest that integration across relevant agencies, including social care and education, could improve service delivery and family experience. However, chronic underinvestment in services has clearly effected capacity (Dowden, 2018) and is compounded by workforce shortages, particularly consultant paediatricians (BACCH, 2019), (see PT3a, skills mix and PT7 training) but we found little evidence at the *macro* level of how this is, or could, be addressed.

Focusing on multi-agency working, structural and organisational barriers include 'poor information and data sharing, lack of resources and additional workloads for services, lack of leadership and clear direction' (The Scottish Government, 2014, p.10). Suggested strategies to address this include: a 'whole life' autism strategy that co-ordinates multi-agency planning; better integrated pathways with ring-fenced funding (albeit appertaining to adults); and a national approach to support children with autism (The Scottish Government, 2014, p.19). Another practical suggestion to promote integration with the voluntary sector is for autism services to use rented rooms for assessment and feedback appointments (Tollerfield and Pearce, submitted).

Finally, Hayes et al. (2018)'s review suggested that despite individual guidelines presenting a coherent assessment process, there is sufficient variation between guidelines to seed confusion; they argue that clinical guidelines for autism diagnosis 'illuminate the process of diagnosis as social rather than straightforwardly clinical, and that judgement is required to consider a number of sometimes contradictory and complex social factors' (Hayes et al., 2018, p.21). The authors do not relate this to the wider context, but presumably if the guidelines have variations, CCGs will interpret differently, which appears to be the case, given the differences in model and skill mix across the UK.

6b. Micro level

Gregory et al. (2013b, p.76) advocates working at the 'network level' to discuss a CYP and family's needs with other professionals in the MDT and across organisations. Working closely with the network improves communication and promotes a consistent approach which can decrease conflict and breakdowns in relationships between different parts of the system; additionally, this makes best use of available skills and increases everyone's skill-set through joint working (Gregory et al., 2013b).

The Scottish Mapping Report (The Scottish Government, 2014, p.10) commented that parents observed a 'lack of co-ordination between different health specialists and between schools and health and education authorities' which was perceived as contributing to the length of the process and lack of support post-diagnosis. Clinicians have also commented on the slow process of multi-agency diagnosis, such that it has been used to justify independent diagnosis (Karim et al., 2014). To improve effectiveness, the Autism ACHIEVE Alliance (2014) report recommends establishing clear pathways and detailing constructive use of time and tools at each stage of the autism diagnostic process. Another way of developing skills through clinical experience (compared to training) is creating split posts or other opportunities to work in different teams within the same geographical area (eg. CDCs or mainstream school services). As such, staff can act as bridges between different parts of the system, aiding understanding and communication (Gregory et al., 2013b).

We found little literature describing how health and education work together, other than endorsing the importance of follow up services communicating directly with the school 'to ensure that teachers understand and can respond to the child's needs' (Calzada et al., 2012, p.240). However, one approach involved an Additional Learning Needs Coordinator (ALNCo, a teacher at the school) who coordinated support for children pre- and post- diagnosis and provided the link between parents, teachers and any other professionals involved in the child's care. The ALNCo, class teachers and other allied staff (including a SALT) were all involved in providing ongoing post-diagnostic support to the child and family (Hurt et al., 2019). However, this is not unique and replicates the role of an Area Inclusion Co-ordinator (InCo) or Special Educational Needs Co-ordinator (SENCo)(Department for Education and Department of Health and Social Care, 2015).

3.7 Programme theory 7 – Training, service evaluation and development

Based on their needs, skills and knowledge for autism diagnostic assessments and working with families, health and community professionals should have access to tailored training, service development and service evaluation.

7a. Training for professionals working with CYP in community settings

The Scottish Strategy for Autism Mapping Report (The Scottish Government, 2014) suggests that training in many organisations is "ad hoc" with gaps, varies across settings/professions and may have low priority given financial constraints; new staff or those in new roles tend to have gaps in their uptake of training; meanwhile, multi-agency training is insufficient.

Several papers identify the importance of effective training in improving the quality and efficiency of Austim diagnostic services (The Scottish Government, 2014, Rutherford et al., 2018). Awareness and training should cover more than professionals working in autism services and include the educational sector (The Scottish Government, 2018) and be geared to the needs of managers as well as front-line staff (The Scottish Government, 2014) through multi-agency training (NICE, 2011). The Scottish Strategy for Autism Mapping Report (The Scottish Government, 2014) suggests autism training should be introduced as part of clinical and professional development for relevant professions, for example, SALTs, social work Learning Disability services and teachers (The Scottish Government, 2018) so that they are better equipped to work with autistic children.

Rutherford et al. (2018) suggests a training framework mapping skills and knowledge for autism diagnostic assessment at different levels. It refers to the NHS Education for Scotland Autism Training framework (2014), which suggests four levels of staff skills and knowledge for autism diagnostic assessment: 1) informed level - for all professionals working with autism in health and social care settings; 2) skilled level - for all staff with direct and/or frequent contact with individuals with autism or those who have a role with high impact on these individuals; 3) enhanced level - for professionals with more regular or intense contact with individuals with autism where their role focuses specifically on the condition and providing interventions, or service managers; and 4) expert practice level – for professionals who have a specialist role in the care, management and support of people with autism. This framework can be used with individuals, organisations or training providers to identify current or future training needs and levels. Detailed descriptions of what is required at each level are outlined in the framework but rather than defining levels specific to profession or position, the level depends on the 'nature, extent and likely impact of daily contact with individuals with ASD' (Rutherford et al., 2018, p.1583). As frontline staff are increasingly likely to come into regular contact with children with autism (and other developmental issues) in generic children's services (e.g. nurseries, play groups), staff need to develop their skills at the enhanced, or even expert level, to facilitate diagnosis (Rutherford et al., 2018).

Multi-agency training for professionals with a targeted and coordinated approach across organisations can facilitate a wide breadth of basic training (The Scottish Government, 2014). For example, Gregory

et al. (2013a) argue that their specialist CAMHS team for children with disabilities, including autism, provides local training to parents, play services, social work teams, CDCs, schools, SALTS and other professionals, which increases the local skill set of those who regularly work with children who may have autism. However, we found no evidence to support the role of tertiary services in training local autism services, as our programme theory had initially predicted.

7b. Training for health professionals working in autism services

Professionals working in ASD services need support including backing for training, funding for a specialist library and practical resources (e.g. camcorder, symbol software and sensory equipment for parents to trial), as well as access to supervision, links with other experienced professionals, and an open team culture of sharing ideas, in order to work with CYP as effectively as possible (Gregory et al., 2013a).

As discussed in PT7a, training programmes need to be tailored to the level of competencies required (i.e. enhanced and expert practice levels) dependent on their role (Rutherford et al., 2018). Rutherford et al. (2016) suggest that training activities could include observing in a specialist autism clinic to develop skills and confidence; 'buddy up' with more experienced staff; regular Continuing Professional Development (CPD) sessions for the team to review training needs; developing an explicit plan for succession planning and training needs (e.g. ADOS); and a national forum to share experiences and knowledge, including people with ASD and their families. As more staff become better trained in, for example, the use of relevant standardised assessment tools, there will be a higher degree of consistency between local and specialist teams. The Autism ACHIEVE Alliance (2014) also suggests carrying out reviews and succession planning of training needs for each service and providing opportunities for CPD so as to develop efficient working and communication.

7c. Service development and evaluation

This section relates to managerial 'training', or audit, evaluation and service development. As autism services have become increasingly 'overburdened', clinicians have been 'unable to engage with service development' (RCPCH, 2017, p.16). Service evaluation is important to check adherence to standards/guidelines but not all services routinely collect relevant data or go beyond basic patient satisfaction audits. Thus, services should maintain or develop efficient systems of collecting information about referrals, waiting times and outcomes to support audit and self-evaluation; a simple way of doing this is to use a guidelines checklist at the front of each patient file (Rutherford et al., 2016). The data (e.g. waiting times) can be collated for senior managers and commissioners to evidence shortcomings in staffing and resources.

In addition to addressing training needs, CPD should be used to review service development, for example, investigating and reviewing the utility of new diagnostic measures and services. Suggestions to help promote service development and embed changes into the care pathway include having one person to lead/champion change; generating research within clinical teams; and encouraging practitioners to co-create contextually sensitive solutions (Rutherford et al., 2016). Practitioners' suggestions would have strong face validity and clinical relevance but still require implementation and evaluation, implying a cyclical process to service development and evaluation, as an ongoing component of CPD. Finally, there are 'experts' within the field, including people with autism, carers and specialist organisations that have considerable knowledge and could support local service development if identified and connected into the process (The Scottish Government, 2014).

3.8 Summary

The chapter presented evidence for each programme theory. The first four theories explored factors related to the referral pathway, from pre-assessment, to triage and referral management, diagnostic assessment, devising a management plan and follow up. The remaining programme theories related to aspects of relevance to the entire care pathway, including working with families; inter-disciplinary and inter-agency working; and training, service evaluation and development. The conceptual map provides a summary of the programme theories and the macro/meso context within which autism diagnostic services operate.

4: DISCUSSION

4.1 Overview and study limitations

The purpose of the RRR is to inform the lines of enquiry in subsequent RE-ASCeD work packages hence the need to develop initial programme theories to test in empirical stages. However, evidence in a realist review is not about providing clear cut facts about whether a particular approach is likely to be effective, but about providing evidence that gives an indication of the possible links between an influencing context, resources, responses of those involved in the service, and outcomes. We explicitly set pragmatic limits on the literature search (Stages 1-2) as RRR allows (Saul et al., 2013) and acknowledge that it cannot be as extensive as a full realist review, given time constraints. However, we were mindful of the need for comprehensiveness without accruing too much extraneous material in the process. Engaging with our expert stakeholders throughout the process enabled an iterative approach to identifying relevant literature. We limited the search to UK literature, because this is pertinent to our research questions, but for the discussion, we have widened our remit to include other literature to elucidate the findings. However, we acknowledge that we may have missed literature from similar health systems that could have informed our programme theories. Despite limits on the search, the synthesis (Stages 3-5) is as extensive as that for a full realist review.

The aim of the discussion in realist terms is to utilise theory in understanding the implications of the findings of the RRR in relation to our overarching research question. We will use Normalisation Process Theory (NPT) (May and Finch, 2009) as a lens to explore the findings and determine how issues in implementation may impact on intended outcomes. NPT assists in explaining the processes by which complex interventions become routinely embedded in healthcare practice. It has been designed for process evaluations and is also suited to comparative studies of complex interventions (May and Finch, 2009). The model identifies and describes factors that may promote or inhibit implementation of an intervention, in this case the autism diagnostic assessment, and provides a basis for assessing the probability that new ways of working will become routinely incorporated, or 'normalised', into clinical practice. In this context, we are using NPT to illuminate the findings, in conjunction with international studies, to draw out key enablers and barriers in implementing ASD diagnosis pathways to deliver high quality and timely autism diagnostic services for CYP with possible autism. NPT uses four main categories to consider implementation processes (May et al., 2018):

i. Coherence: how stakeholders individually and collectively understand autism, the diagnostic pathway and support services. For example, how parents view the purpose of the service compared to clinicians, potentially leading to unintended outcomes, such as parental dissatisfaction.

ii. Cognitive participation: how different stakeholders 'buy-in' to the service. For example, if the service model requires a different way of working that practitioners are not familiar with, then the model may face challenges in delivery.

iii. Collective action: the actual work required to deliver the autism diagnostic care pathway. For example, if the model is too resource-intensive, in the context of meeting the clinical need, it may not be feasible to deliver the service.

iv. Reflexive monitoring: how stakeholders individually and collectively evaluate the service. For example, commissioners, providers and parents may define positive outcomes in different ways.

4.2 Coherence: individual and shared understanding of autism and associated diagnostic services

4.2.1 Training needs and competency

Programme theory 1b explored the evidence around how best to enable frontline health and education professionals to be cognisant of autism and referral pathways. The findings were consistent in supporting the need for greater autism awareness and training in primary care and educational settings, alongside training in how, when and who to refer (The Scottish Government, 2014, Rutherford et al., 2016, O'Reilly et al., 2017, Unigwe et al., 2017, Crane et al., 2018, Dowden, 2018). This should include awareness of atypical and more complex presentations which are often subject to delays in recognition, for example the child who also has ADHD (Brett et al., 2016, Kentrou et al., 2019).

Our Expert stakeholders also suggested that a shared understanding of autism and the local referral pathway could be enabled through early familiarisation of the impact and experience of families living with autism and examples of local initiatives were given such as the Time for Autism project. This project is based on a Time for Dementia programme which stemmed from concerns that training for healthcare professionals was overly focused on acute care, reinforced by the nature of short-term clinical placements, and this precluded students from developing an understanding of long-term conditions which requires a different skill set (Banerjee et al., 2017). The programme is an interdisciplinary educational programme whereby medical, nursing and paramedic students visit a person with dementia and their family for 2 hours, every three months, for two years; they follow a semi-structured interaction guide focusing on experiences of illness and services and complete reflective appraisals (Banerjee et al., 2017). Evaluations suggest that for families, involvement in the

programme can be therapeutic (Cashin et al., 2019) and for student health professionals the programme can promote understanding of the perspective of people with dementia (Grosvenor et al., 2017).

Similarly, Time for Autism is intended to become a mandatory part of the fourth year of the medical curriculum at Brighton and Sussex Medial School. It will include classroom-based learning from specialists in the field (including those with autism) alongside visiting a family who has a young person on the autism spectrum and visiting them three times over the course of a year (commencing September 2021). The intention is that future doctors should be equipped 'with the appropriate skills, knowledge, empathy and understanding to provide good quality care' to those with autism and their families (Brighton & Sussex Medical School, 2020) although the programme has yet to be implemented and evaluated. There is already a funded plan in place, including for a research fellow to evaluate this programme once it starts.

Linked to understanding the referral criteria and diagnostic pathway (PT1) are the training needs of professionals working with CYP in community settings (PT7a) and autism services (PT7b). The evidence indicated that all health and community professionals working with CYP should have access to tailored training, service development and service evaluation and that training should be tailored to the level of competencies required dependent on their role (Rutherford et al., 2018). However, this raises two issues: first, how to judge professional competence and second, how to ensure training is needs-led rather than, as currently, often aligned with promotion criteria. Job roles within the NHS are 'scored' against fixed criteria and matched with a specific pay band depending on how high that score is (Royal College of Nursing, 2020). For example, specialist nurses wanting a Band 8 position are required to hold a Masters' degree yet there is no degree specific to their role in autism services. It may be more useful to develop an autism specific training package similar to one developed by, for example, Health Education England (2018) to address dementia education. This training package is divided into three tiers, based on a dementia training standards framework that describes core skills and knowledge for each tier: firstly, awareness, which everyone should have; secondly, basic skills, relevant to all staff in settings where people with dementia are likely to appear (similar to PT7a) and; thirdly, leadership, for managers/service leads in a position to enhance the knowledge, skills and attitudes for key staff (experts) working in dementia care.

Although we did not find published evidence, our expert stakeholder group suggested that having a local tertiary service or centre of excellence could provide a catalyst for local training initiatives, with an implicit impact on a shared understanding of autism and associated regional services. The example of paediatric services for epilepsy was cited as a potential route to follow. Following an acknowledgement of diagnostic and prescribing challenges within epilepsy services for children

(Kitson and Shorvon, 2000), the British Paediatric Neurology Association spearheaded a series of workshop-based paediatric epilepsy training courses based on published clinical guidance that could be delivered by a suitably trained and experienced faculty. Three levels of Paediatric Epilepsy Training course were designed, aimed at a variety of health professionals from trainee paediatricians, Epilepsy Specialist Nurses and children's nurses, general and community paediatricians, through to tertiary paediatric neurologists (Kirkpatrick et al., 2014). This programme of training has been seen as a major contribution to the upskilling of paediatricians and nurses working within CYP's epilepsy pathways, and enabled an expansion of CYP's epilepsy networks throughout the UK (Kirkpatrick et al., 2014). Such a model, whether delivered regionally, or nationally, should be possible around autism diagnosis and management, and its associated comorbidities and differential diagnosis.

Gordon-Lipkin et al. (2016) provides useful examples from the USA of innovative approaches to promote early identification of autism and reduce waiting lists. For example, the Vanderbilt Kennedy Centre (Nashville, USA) developed a programme intending to reducing waiting times in the state of Tennessee by training primary care paediatricians to use a validated screening tool for autism, called STAT (Screening Tool for Autism in Toddlers and Young Children) (Vanderbilt Kennedy Centre, 2020). It was designed as a Level 2 screener for use in referral settings, to identify children who are at risk for autism where there are already developmental concerns and was designed for use 'by a wide range of community professionals - not only to increase the rates of early detection, but also to enhance community awareness of the early signs of autism' (Vanderbilt Kennedy Centre, 2020). The assessment evaluates toddlers (24-35 months), takes about 20-minutes and uses twelve interactive items based on social-communication behaviours evidenced to differentiate two-year olds with and without autism. The tool has been validated to identify to correctly identify 75-90% children with autism and can be carried out during routine 18- and 24-month paediatric visits (Stone et al., 2004, Gordon-Lipkin et al., 2016). Our expert stakeholders suggested the A.L.A.R.M (Autism is prevalent, Listen to parents, Act early, Refer, Monitor) Surveillance and Screening Algorithm in the USA, which was developed to establish standard practices among physicians, to simplify the screening process, and to ensure that all children receive routine and appropriate screenings and timely interventions(Centers for Disease Control and prevention (CDC), 2019). However, in the UK, screening tools have not been widely adopted in view of concerns over low sensitivity and specificity because, as NICE (2011) points out:

- a positive score on tools to identify an increased likelihood of autism may support a decision to refer but can also be for reasons other than autism, meaning that some CYP may be referred unnecessarily and

- a negative score does not rule out autism, meaning that some families may be falsely reassured that their child does not have autism. Using the example of the STAT (above), if only 75% are correctly identified as having autism (sensitivity) then 25% (1 in 4) who in reality have autism remain undetected. Similarly, another study found a sensitivity of only 52% and a specificity of 84% when looking at 10 year olds who had been screened at 2 years old using M-CHAT (Kim et al., 2016). This is further compounded by the risk that CYP with more subtle forms of autism (e.g. those previously described as having Asperger's syndrome) may not be detected because they do not necessarily present with the symptoms these early childhood screening tools are designed to detect, but because parents are falsely reassured they do not seek help when symptoms become more evident as the child reaches school age (Kim et al., 2016).

The Vanderbilt Kennedy Centre (Swanson et al., 2014) also initiated a training programme with the aim of developing a network of primary care paediatricians who are able to identify and refer children for specialist assessment. Training involved a 2-day workshop focusing on: administration and scoring of the STAT and a Modified Checklist for Autism in Toddlers (M-CHAT); carrying out a parent interview; integrating the information to generate a diagnostic impression; explaining the results to families; and payment issues relevant to the USA healthcare system. Although paediatricians identified significant barriers related to time and financial pressure, the majority of participants reported substantial changes related to primary diagnostic assessment practices and an 85% increase in diagnostic identification of children with autism within their practice setting (Swanson et al., 2014). However, the study was unable to elicit 'whether practitioners were in fact providing diagnoses in place of or prior to expert evaluation' (Swanson et al., 2014, p.5). Therefore, there is a risk that brief diagnostic models could result in misdiagnosis but this has to be balanced against the benefits of early identification and access to services for a greater number of young children. Additionally, most of the paediatricians participating were very experienced, highly motivated and were already routinely conducting brief ASD screening which raises questions around the baseline level of skill and motivation needed to achieve accurate diagnosis and sustained change in practice (Swanson et al., 2014). This also draws attention to the difference between providing training to deliver a specific diagnostic tool and comprehensive training to understand autism, differential diagnosis and comorbidities. Additionally, the high costs of using standardised tools, such as ADOS, and the costs of training professionals to use them, potentially limits the number of proficient providers, particularly in resources-limited settings, highlighting the need for open access tools (Gordon-Lipkin et al., 2016).

Another example of supporting primary care constitutes a community-based Autism Liaison and Treatment training programme (Arkansas, USA) to establish 'mini-teams' of one community

paediatrician and one SALT able to provide diagnostic developmental assessments. Of note, community paediatricians in USA, Canada and Australia are generalists working at primary care community level, whereas in the UK community paediatricians are specialists in neurodevelopment, working in secondary (or tertiary) level care. The emphasis of this programme was on connecting clinicians to representatives in appropriate early intervention services in order to facilitate network building and communication within the professional community that serves these children (Gordon-Lipkin et al., 2016).

Penner et al. (2017)'s Canadian study focuses on the second stage of referral, community general paediatricians who receive referrals from primary care practitioners, and then complete their own assessment and decide whether to refer to a more specialised provider. The context is that of paediatricians working in isolated and remote communities with access to specialists that are centralised in, for example, Toronto. Although only eleven paediatricians participated, the study resonates with aspects of our programme theories, including training needs. The participants were trained to work with developmental issues in tertiary care settings but these did not always translate to the resources and requirements of community practice. Additionally, diagnostic decision-making was influenced by a variety of factors relating to the child/family (e.g. language/culture), the clinician (e.g. beliefs about autism, section 4.2.2), and contextual/systems factors (e.g. remuneration). Communicating the diagnosis and managing follow-up also influenced diagnostic decision-making, for example, gauging the family's reaction to a possible diagnosis and lack of follow-up services (Penner et al., 2017).

The Australian guidelines (Whitehouse et al., 2018, p.25) clearly state what competencies and training are required for primary healthcare providers and for those who provide specialist assessment. The guidelines recommend that primary healthcare providers (including community nurses, midwives, AHPs, pharmacists and dentists) have received:

> *'formal professional training in typical child development and the signs and/or symptoms of common neurodevelopmental and behavioural conditions, including those associated with ASD, as well as common co-occurring and differential diagnosis conditions'*

and additional training if required to administer clinical assessments as part of the referral process. For clinicians involved in autism assessment, in addition to local clinical training, clinical networks should facilitate the training of new team members to develop 'expertise through peer observation, peer supervision and peer mentoring', supplemented by formal training courses and/or further qualifications (Whitehouse et al., 2018, p.17). We only found one example, Peterborough's Integrated Neurodevelopmental Team (Autistica, 2019), where the link between training, competencies and

diagnostic assessment is explicit: 'professionals are selected to undertake the assessment based on their competencies and child's needs rather than professional background' (Male et al., 2020, p.2). However, our expert stakeholders cautioned that while the literature demonstrates interesting possibilities there is little evidence to endorse their implementation, or transferability, from other countries to the UK health, social care and educational context, and that other pertinent literature may not have been captured within the remit of this RRR.

4.2.2 Inter-professional working and paradigmatic dissonance

Programme theory 6 identified the importance of inter-agency working to develop shared knowledge that can be embedded into local services to build capacity, improve parent/CYP satisfaction and support service planning both locally and nationally. However, many structural and organisational barriers to inter-agency working were identified, including tensions between different views of autism (Abbott et al., 2013, Dowden, 2018); lack of cross-over between health and education (Hurt et al 2019); variation between guidelines (Hayes et al., 2018, Penner et al., 2018a); and commissioning and funding arrangements (The Scottish Government, 2014, Male et al., 2020). Underpinning these barriers were disparate views on what autism actually constitutes and how to interpret it. Furthermore, Hayes et al. (2018, p.2) highlight that 'assessments are contextual and inter-relational and symptoms may change according to context or interpersonal relationship' which can result in contradictory findings.

Penner et al. (2017)'s qualitative study noted that community paediatricians contrasted autism against 'medical' conditions that were more clearly aligned with their medical role. Given the Canadian healthcare system, there was the added dimension of an intersection between the medical role and remuneration, with other conditions being better compensated because they could be assessed in shorter intervals. Our expert stakeholders also commented on paradigm tensions between doctors and educational psychologists. For example, as a co-occurring condition dyslexia is important in terms of differential diagnosis but educational psychologists often perceive this as solely within their remit. Whether this contributes to the lack of cross-over between health and education is unclear but it does reflect arguments around the socially constructed and culturally specific nature of autism (Begon and Billington, 2019) and how it is understood and managed differently in different parts of the world (O'Dell et al., 2016).

Two key perspectives dominate: a medical view of autism based on a deficit model and constructed from an 'outsider' perspective contrasting with an asset model, based on neurodiversity and having evolved from people with autism themselves. Although Begon and Billington (2019, p.186) acknowledge that both models are 'conceptually incomplete', they suggest that educational

psychologists should adhere to the neurodiversity model, focusing on autism as neurological difference rather than deficit (Silverman, 2015, O'Dell et al., 2016, Begon and Billington, 2019). This is reflected in the role of educational psychologists, working from a 'consultation framework' to support CYP 'to retain a sense of the individual, as someone with a distinctive profile of strengths and needs and around whom a bespoke and multi-faceted package of support must be tailored' (Begon and Billington, 2019, p.190-191). This brings into focus the dilemma that CYP have traditionally needed diagnosis to access services (Penner et al., 2018a), despite more recent (Australian) guidance advocating for needs-led provision:

> *It is critical that a client's needs, not the presence or absence of a diagnostic label, are used to determine eligibility and prioritisation of access to intervention and support services'* (Whitehouse et al., 2018, p.4).

Similarly, the NICE quality standards (2014, p.15) emphasises the need to develop a profile of the CYP's 'strengths, skills, impairments and needs' including intellectual ability, speech, motor, sensory, adaptive behaviour (e.g. sleep, eating) mental and neurodevelopmental health', including the need to assess for co-existing physical and mental health problems including genetic, cerebral palsy and epilepsy, and to develop a personalised plan in partnership with family, and other agencies (e.g. school). This may include contributing to an Education Health and Care Needs plan.

Although educational psychologists may want to challenge medical doctrine, they have to rely on medical diagnosis (as do parents) to receive support and funding (Hurt et al., 2019). Our stakeholders commented how this can manifest in tension between parents, schools and clinicians, with pressure on clinicians to diagnose, including from some SENCOs, because it is regarded as essential to access educational psychology and support services.

4.3 Stakeholder 'buy-in': national policy, local services and integrated working

4.3.1 National policies versus local priorities

Different perspectives of autism at the micro level (clinician, parent, teacher) were reflected at the meso level, in terms of inter-professional and inter-agency working (PT 6). This was complicated by a dichotomy of centrally imposed policies (meso level) set against local context and buy-in. Variation often reflects historical service development, for example, our expert stakeholders cited West Sussex, where there is a single joint commissioning unit, although several CCGs. Each CDC, three of which are now in the same health trust, have subtle differences in practice which reflect a history of working in different organisations, differences in access to CAMHS and the personal practices of clinicians leading the autism service. In Wales, one explanation for the 'ad hoc' arrangements between health and

education was that the organisation of the health service (managed by seven Health Boards) was different to the organisation of education services (managed by 22 local authorities), and each organisation had its own practices (Hurt et al., 2019). Similarly, the drive for Integrated Autism Service (IAS) for adults in Wales experienced delays in some areas 'caused by difficulties securing local "ownership" of the national IAS model' (Holtom and Lloyd-Jones, 2019, p.9), although early reports were positive in terms of improving capacity (in some areas only) and the user experience. Linking integration with training, the IAS:

> *provided a focal point for consolidating autism expertise and a resource for joint working, consultancy, advice and training to raise awareness and upskill other services, most notably mental health services* (Holtom and Lloyd-Jones, 2019, p.10).

Penner et al. (2019) compared Canadian and UK government policies that govern autism diagnostic practice. Unlike in the UK, where service delivery is directed at a national level, the federal government in Canada provides funding through health and social transfers to the Canadian provinces and territories, which have jurisdictional control over how health and social services are delivered. The review found considerable variability in how assessments were carried out, including what tools were used, who carried out the assessment and provision of intervention. While diagnosis was provided by health services, interventions were provided by several ministries including health, education, family/children, or community and social services. This division between assessment and intervention:

> *creates a potential disconnect for resource considerations such as advocating for resource-intensive diagnostic assessments without considering the impact on service delivery, or conversely, implementing lax diagnostic requirements without consideration of the impact of false-positive cases on wait times and costs* (Penner et al., 2019, p.133).

This highlights a lack of integration between diagnostic assessment and intervention strategies and that the interplay between them 'have not been fully elucidated beyond simply determining eligibility for costly interventions' (Penner et al., 2019, p.132).

4.3.2 Integrating services

Allard et al. (2019)'s comprehensive report on integrating services for disabled children highlights that integration is not a homogenous approach, for example, care can be integrated at different levels, around the individual, across multiple services, or system-wide. However, strategic leadership with a clear strategic vision is 'the single most important factor in enabling or hindering joint working and integration at local level' and is reliant on 'local leaders' power to set strategy, influence organisational culture and support initiatives that enable integration' (Allard et al., 2019, p.28).

It is clear that structural and cultural differences between different services (and across primary and secondary care) can impede the delivery of integrated services in whatever setting (Coughlan et al., 2020). Conversely, there is some evidence that aligning services and funding improves outcomes in relation to waiting times, duplication of assessments and associated costs, and an improved experience for parents and CYP (Galliver et al., 2017, Hayes et al., 2018, Rutherford et al., 2018, Autistica, 2019, Hurt et al., 2019, Male et al., 2020). However, aligning and integrating services relies on 'buy-in' at all levels, including commissioning and organisational, but ultimately rests on the engagement of clinicians to deliver the service (Coughlan et al., 2020). Our stakeholders cited examples of the reverse, where clinicians were committed to joint working (micro level) but the initiative failed due to reservations at the management (meso) level. Male et al. (2020) suggest that if all service providers work within a single organisation accountable for the delivery of an integrated service, it is likely to reduce the pressure on each part of the team (e.g. CAMHS or CDC) to deliver diagnostic assessments for other parts of the team. This would also enable an approach where the clinicians required for assessment would be 'based on the competencies required to address the specific presentation and needs of the child, rather than which service they are referred to' (Male et al., 2020, p.3), again highlighting competencies. The user experience might also be improved if the neurodevelopmental pathway is aligned with Early Help Pathways, and pre- and post-diagnosis parent training programmes, commissioned jointly by the Local Authority and NHS (Male et al., 2020).

Finally, Wolfe et al. (2020) carried out a systematic review and meta-analysis (1990-2018) to assess the effects of integrated care on child health, healthcare quality, use of services and costs for CYP with ongoing conditions. While the eighteen included studies were diverse in methodology and setting, and there is no validated measure of extent of integration, Wolfe et al. (2020, p.9) concluded that 'integrated models of care may deliver some important improvements in QoL [quality of life] for CYP with ongoing health conditions and potentially save money'. Importantly, they were unable to discern which children benefited most and suggested that socio-economic factors may impact on outcomes. This appears logical but is perhaps of particular relevance for CYP with autism, given the social/cognitive and educational aspects of the condition (section 4.2.2).

4.4 Collective action: doing the work

4.4.1 A flexible approach to diagnostic assessment

This section links mainly with programme theory 3, the diagnostic model and skill mix. There was no 'one' approach or model to diagnosis but MDT assessment is strongly supported by most practice guidelines (Hayes et al., 2018). However, there are occasions when a single experienced clinician could make a diagnosis with the caveat that a comprehensive profile still requires a multidisciplinary approach (Hayes et al., 2018). However, NICE guidelines (2011) strongly recommend MDT assessment,

reflecting concerns over reliability and increased potential for individual bias for, or against diagnosis, with single practitioner assessment, echoed by research showing disagreement in diagnostic conclusions between single practitioner and multidisciplinary assessment in around 7-20% of cases (Swanson et al., 2014, Hausman-Kedem et al., 2018).

Hayes et al. (2018) noted that none of the guidelines address how clinicians actually reach consensus within a multi-disciplinary context and our expert stakeholders raised issues around one profession trusting another's clinical judgement**,** for example, SALTs having reservations about nurses taking on their role in observational assessment. Perhaps this relates not solely to trust but also to *evidencing* professional competencies in a consistent manner across disciplines.

Variation across guidelines may reflect the heterogeneity in clinical presentation associated with autism and that a 'one-size-fits-all' approach to diagnostic assessment lacks sufficient flexibility to accommodate this (Penner et al., 2018a, p.525). For example, CYP with a severe presentation of autism may not require multiple assessors and diagnostic tools to confirm a diagnosis with acceptable accuracy and 'inflexible recommendations and requirements for assessment may add inefficiency to an already strained system' (Penner et al., 2018a, p.526). Therefore, there may be a trade-off between carrying out comprehensive assessments for all CYP with possible autism and 'providing a more streamlined approach that is tailored to the child's presentation' (Penner et al., 2018a, p.526).

St Helen's Neurodevelopmental Pathway (North West Boroughs Healthcare NHS Foundation Trust, 2020) is an example of a service that carries out multi-agency assessment for CYP with complex neurodevelopmental difficulties and has built flexibility into the process. Pre-referral, the service provides training for those who are likely to work with CYP, support/training for families (eg. Positive Parenting Programme), and access to universal services. Referral is via a single point of access and triaged by the MDT. Referrals that require a single agency assessment are referred to the appropriate service and closed to St Helen's. For CYP that are accepted, referrals are made to initiate multi-agency specialist assessment with the emphasis on description of need, rather than diagnosis. This leads to an inter-disciplinary discussion of the findings, including representatives from health, educational psychology, local authority/social care and the development of a multi-agency support plan. This is formally fed back to the family, although it is unclear how they are involved in developing the management plan. CYP with no formal diagnosis are able to access universal or targeted services dependent on need and those who receive a mental health diagnosis are managed by Barnardo's or CAMHS.

Somewhat similar to Rutherford et al. (2018)'s abbreviated and complex pathway, the Australian guidelines (Whitehouse et al., 2018) advocate a flexible and progressive approach, whereby additional

clinical investigations are based on the clinical complexity of the individual. The model starts with an *assessment of functioning* and needs (medical and/or AHP) combined with *medical examination* relevant to neurodevelopment disorders; if diagnostic evaluation is supported, this is followed by a *single clinician diagnostic assessment*, carried out by a doctor or psychologist; and if high diagnostic confidence is not achieved, the final stage is a *consensus team diagnostic evaluation* which builds on the previous assessments, and includes AHPs and medical practitioners. However, it is not essential to apply the stages consecutively and implementation can be adapted depending on the clinical history of the individual (Whitehouse et al., 2018). Perhaps this illustrates the complexity of autism diagnostic assessment and how services need to be designed to take account of the complexity associated with certain presentations and family situations. For example, Brett et al. (2016) investigated phenotypic factors associated with age at ASD diagnosis in the UK (n=2134, age 2–18 years). Factors associated with earlier age of diagnosis were autism diagnosis (compared with other ASD), language regression, language delay, lower socioeconomic status, and greater degree of support required. They also found that the median age for diagnosis had not decreased in the UK in the last decade, indicating that there are certain groups for whom clinicians might develop additional strategies to reduce the age of diagnosis in the future.

4.4.2 Engaging parents and CYP as co-experts

Ashcraft et al. (2019)'s systematic review aimed to identify the antecedents and consequences of parental empowerment in paediatric healthcare settings. Most articles (32 out of 44) had no formal definition but defined empowerment, at least partly, by anticipated outcomes such as symptom management. However, the parent-provider relationship was clearly associated with parental empowerment and manifested in, for example, attentive communication/being heard and trust between parent and clinicians (both akin to PT 1a), and family perception of being an equal team member (Ashcraft et al., 2019) (similar to PT 5a, parents as co-experts). In contrast, interactions with providers who were perceived as insensitive or poor communicators appeared to contribute to feelings of disempowerment. Equally important was engaging parents in the actual process through, for example, shared decision-making and goal setting. Receiving information and support were associated with empowerment and conversely, lack of information, being overwhelmed with information and advice, or having unanswered questions were associated with disempowerment. Overall, no link was identified between parent empowerment and healthcare utilisation. Allard et al. (2019, p.40) also advocate participatory approaches with parents and CYP, linking with integration, by involving families in decision-making to:

> *support integration at an individual level by uniting professionals around holistic ambitions and outcomes. This should also be true at a strategic level, where effective participation and*

co-production should have the effect of uniting different agencies around what matters to families.

4.4.3 Telehealth and telemedicine

The literature tends to use the terms telehealth, telemedicine and digital technology (programme theory 3c) interchangeably but there is emerging research on applying telemedicine for CYP with autism. Given the pace of improvements to technology, increasing referral rates, and the current context of the Covid-19 pandemic, this is particularly relevant. For example, autism evaluations performed through video conferences between a family and the MDT may be an accurate means of assessment (Gordon-Lipkin et al., 2016) and a potential means of initially screening referrals. Other examples include designing a diagnostic tool using home videos recorded by parents; local assessments which are presented to a university-based medical centre team that makes the final diagnostic assessment and provides recommendations remotely; and providing peer-to-peer training or advice (Gordon-Lipkin et al., 2016) which could help support and train clinicians in remote/rural communities.

The advantages of telehealth include the ability to work with families living in rural or remote locations with clinicians who have expertise that may not be available locally, and to facilitate the assessment of individuals who may be reluctant to attend a clinical setting due to, for example, sensory symptoms (Whitehouse et al., 2018). However, there is little data to evidence that telehealth leads to a valid diagnosis and there are other limitations, particularly in the breadth and detail of information that can be gleaned from an individual and the conclusions that can be drawn from an assessment using this 'restricted' communication method (Whitehouse et al., 2018, p.23).

Indeed, the challenge created by Covid-19 and the need for social distancing may well drive the introduction of further digital solutions to autism assessment, where previously direct observation of, and interaction with the CYP (NICE, 2014) has been considered essential. However, our expert stakeholders raised two key issues. First, the issue of digital poverty, that not all families have access to technology, and that children do not always comply with instructions to play in front of a camera. Second, that although remote observations/ consultations (in the context of covid-19) appear to have largely positive feedback from parents this stems from necessity rather than diagnostic validity, confidence in the findings and relationship with the clinical team. The research evidence is at an early stage and more evaluation is needed to explore further sensitivity and specificity for best estimate clinical diagnosis and use in real practices with the full range of children referred.

4.5 Appraisal: service development and evaluation

4.5.1 Integration and quality improvement

Programme theory 7c highlighted the importance of quality improvement in service development and evaluation. Coughlan et al. (2020, p.2) draws together quality improvement (QI) and integration, asserting that QI 'may form part of a wider strategy supporting integration of care at local, regional or national levels, or stem from grassroots initiatives' at the local level. This combination of top-down and bottom-up approaches suggests that there is no 'ideal' approach and that it is possible to combine approaches to drive change, although sustainable change is dependent on the buy-in of all stakeholders, at all levels, including service users.

Coughlan et al. (2020) outlined general principles supporting improvement across whole systems including stakeholder engagement; a co-ordinated strategy; effective and accountable leadership which includes flattening hierarchies to encourage staff feedback; and maintaining staff involvement through inter-professional education, building relationships within and between clinical teams, and training and up-skilling healthcare professionals. Furthermore, meaningful patient engagement should be integral to service development and involve vulnerable patients, or their advocates, to avoid intensifying existing health inequalities. Coughlan et al. (2020) sees a role for patients in all stages of QI from design to dissemination, including identifying outcome measures that matter to them (Coughlan et al., 2020).

Although not related to diagnostic assessment, the Royal Manchester Children's Hospital, a tertiary provider of paediatric healthcare, carried out a process of continuous service improvement. The hospital serves a large proportion of CYP with autism (Kennedy et al., 2016) and wanted to improve the care pathway. Methods included holding stakeholder events to identify barriers to hospital attendance (e.g. sensory overload) and departmental audits. This developed into an ongoing training programme to increase awareness and help healthcare staff deliver care more creatively, events with parents and CYP to share their experiences, and new ways of working as staff gained confidence. Alongside this, the induction process for new staff included attention to autism-friendly working practices, and undergraduate students (AHPs and medics) on placement also had similar training in autism awareness (Kennedy et al., 2016).

Lastly, the Welsh guidelines for adults (Holtom and Lloyd-Jones, 2019) suggest that there needs to be a mechanism to monitor compliance with national standards, reliable methods of measuring improvement and reporting outcomes, and a benchmarking of outcomes across services. This should allow for and explore differences in outcomes, given the likely impact of contextual issues such as resources and staffing.

4.5.2 Data collection and sharing

Related to QI, but at the population level, is the need for good quality data to facilitate a whole system approach (Allard et al., 2019). The Australian guidelines (Whitehouse et al., 2018) recommend a national register to facilitate population-level monitoring of autism prevalence, provide a baseline for longitudinal research and help inform national policy. In the UK, the Database of Children with ASD Living in the North East of England covers six areas around Newcastle (McConachie et al., 2009), whilst a partnership between Newcastle University and the charity Autistica manage the Autism Spectrum Database-UK (ASD-UK) covering the rest of the UK (Autistica, 2020). While a national register should enable stakeholders to 'respond together as a system', the current approach to data collection appears somewhat fragmented making it difficult to measure progress or outcomes 'in a meaningful way' (Allard et al., 2019, p.34).

At individual level, Allard et al. (2019) found that all clinicians supported better data and information-sharing. If different organisations are able share information related to a particular individual, this promotes effective joint-working and integrated care. However, information sharing between different services and agencies is hindered by data 'being held in multiple places, incompatible IT systems and differing governance and security arrangements between agencies' (Allard et al., 2019, p.35). Additionally, our expert stakeholders recommended collecting local and national data around service demand (e.g. referral rates) *across* child development and mental health services as current national data is from mental health services but not CDCs, and sources like the SEN register and GP databases are likely to under-represent numbers significantly.

4.6 Summary

We used NPT to elucidate key dilemmas and areas for further exploration. Key concerns around different understandings of the autism diagnostic pathway and support services included how to judge professional competence and how to ensure training is needs-led. In terms of how different stakeholders 'buy-in' to the service, perspectives of autism at the micro level (clinician, parent, teacher) were reflected at the meso level, in terms of inter-professional and inter-agency working; strong leadership with clear strategic vision appeared to be the most effective approach to address barriers to integrated working. With regards 'doing the work', guidelines varied, as did their interpretation at the local level, but this may reflect the heterogeneity in clinical presentation associated with autism and the need for a flexible approach. Finally, service evaluation needs good quality data to facilitate a whole system approach to service development.

CONCLUSION

Evidence in a realist review is not about providing 'facts' about whether a particular approach is likely to be effective, but about providing evidence that gives an indication of the possible links between an influencing context, resources, responses of those involved in the service, and outcomes. Many of the issues identified in the RRR relate to standards of practice recommended by NICE (2011, 2014). However, there appears to be a gap between the guidelines and local interpretation, exacerbated by demand outstripping capacity and resourcing constraints. As previously noted, there may be a trade-off between carrying out comprehensive assessments for all CYP with possible autism and 'providing a more streamlined approach that is tailored to the child's presentation' (Penner et al., 2018a, p.526). This mirrors feedback from our expert stakeholders – that there may need to be a discussion around the potential to increase investment in service delivery to enable high quality and timely approach versus the potential challenges associated with accepting lower quality and less timely diagnostic assessment.

The programme theories helped identify six key areas that would benefit from further exploration (Table 7). Work package 4, as part of the wider Re-ASCed study, will provide the opportunity to explore some of these areas further, to some extent depending on the case studies selected.

Table 7 Areas for further exploration

Area for further exploration	Related programme theories
1. Evaluation of training and support materials available for:	
a) Non-specialist staff	1b. Frontline health and education professionals are cognisant of ASD and referral pathways 7a. Training for professionals working with CYP in community settings
b) Parents and CYP accessing the service.	5b. Supporting parents/carers
2. Evaluation of training packages for those working in autism services to upskill, and evaluation of the impact on workforce shortages	3a. Model & skills mix 7b. Training for health professionals working in autism services 7c. Service development and evaluation
3. Evaluation of asset-based approaches to diagnosis and support in autism	3a. Model & skills mix 4a. Diagnostic feedback to parents and CYP
4. Evaluation of the barriers and facilitators to comprehensive needs-led diagnostic assessment	3a. Model & skills mix 5a. Parents as co-experts in the diagnostic process

	5b. Supporting parents/carers
5. Evaluation of approaches to integrating services dealing with autism	3a. Model & skills mix 6a. Inter-agency working: macro-meso level 6b. Inter-agency working: micro level
6. Evaluation of the use of digital technology in diagnostic assessment	3c. Digital technology

The six areas worthy of further exploration are:

1. **Evaluation of training and support materials available for a) non-specialist staff and b) parents and CYP accessing the service.**

   Opinions were divided about the remit of GPs in autism diagnosis, ranging from awareness of local services and signposting to taking on a specialist role (Unigwe et al., 2017). However, there was evidence that GPs (and other healthcare/educational professionals) would benefit from a better understanding of autism and when/how to refer for autism diagnostic assessment (programme theory, PT, 1b). The evidence also indicated that anyone working with CYP in community settings would benefit from basic autism awareness training (PT7a), but we found only one example (Gregory et al., 2013a) of a service working with parents *and* a wide range of services (e.g. social workers, teachers) to accomplish this. Similarly, there was limited evidence appertaining to training/support resources for parents and CYP accessing CDC/CAMHS/integrated services (PT5b).

2. **Evaluation of training packages for those working in autism services to upskill, and evaluation of the impact on workforce shortages.**

   The literature related to training packages for those working in autism services (7b) was limited and only the Vanderbilt Research Centre in the USA provided details of specific training packages. However, Tollerfield and Pearce (submitted), in the UK, made the link between upskilling available staff (e.g. SALTs) to alleviate the shortfall of other disciplines (e.g. child psychiatrists) and reduce costs (given the pay differential between AHPs and doctors). There was also limited evidence linking training to wider service evaluation and development (7c) and how this could address workforce shortages. We have provided examples of training programmes that could be adapted or developed for those working in autism services.

3. **Evaluation of asset-based approaches to diagnosis and support in autism**

   Some services take an assets-based approach (PT3a), assessing strengths as well as difficulties, and parents valued this especially when receiving feedback from the diagnostic assessment (PT4a).

However, only one study (Tollerfield and Pearce, submitted) gave a clear description of how this manifested in the diagnostic process and the benefits for CYP and their parents.

4. **Evaluation of the barriers and facilitators to comprehensive needs-led diagnostic assessment**

The NICE guidelines (2011) endorse a needs-led, rather than yes/no, approach to diagnostic assessment. This brings into focus the dilemma that CYP have traditionally needed diagnosis to access services (Penner et al., 2018a) despite more recent advocates for needs-led provision (Whitehouse et al., 2018, Lord, 2020). Parents clearly want appropriate support (PT5b) based on need and regardless of the diagnostic label (Crane et al., 2016) but from the current literature it was not possible to ascertain the barriers or facilitators to providing a comprehensive needs-led service, particularly for CYP with co-occurring conditions. This would allow services to address barriers and capitalise on facilitators alongside evaluating potential (including economic) gains.

5. **Evaluation of approaches to integrating services dealing with autism**

We found limited evaluation of approaches to integration across disciplines and organisations involved in the diagnostic process. One recent systematic review (Wolfe et al., 2020) suggested that integrated models of care may deliver improvements in quality of life of CYP, and possibly save money, but was unable to discern which children benefited most, how or why, or the impact that socio-economic (or other) factors may have on outcomes. The literature also identified difficulties around co-ordination of support pre- and post-diagnosis and who is best placed to help parents navigate the system (Hurt et al., 2019), which would presumably be less of an issue were services integrated.

6. **Evaluation of the use of digital technology in diagnostic assessment**

There was limited literature around the use of digital technology (PT3c), it is in the early stages of evaluation, was not integrated with literature discussing the model or approach to diagnostic assessment (PT3a) and had disparate aims:

- 'Remote' observational assessments of CYP combined with interviewing parents (Juarez et al 2018).
- Using mobile technology as an opportunity to collect data pre-assessment (Tryfona et al., 2016).
- Specific educational games (as apps) that can potentially identify the risk of autism (Tryfona et al., 2016).
- An automated story that scores emotional cognition in children (Jordan et al., 2017).

Three months into the current Covid-19 pandemic, we found further examples of clinicians using remote technology to support diagnostic assessment of mainly younger children in their own

homes, with evidently positive feedback from parents (Lord, 2020). However, approaches were implemented rapidly and on the basis of necessity rather than diagnostic validity. Further evidence is needed about the acceptability, feasibility and utility of use within the NHS and related services (e.g. school setting).

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Appendix A. Database Search Strategies

Number of databases searched: 7

Medline (Ovid)
Embase (Ovid)
PsycINFO (Ovid)
Social Policy & Practice (Ovid)
CINAHL Plus (EBSCO)
Cochrane Library
Web of Science (Clarivate)

Search limits: 2011-current; English language; UK only

Literature search strategy used for **Medline** (search run on 25.11.19) is attached below. Additional search strategies available from the authors.

1	exp Autism Spectrum Disorder/di, ep, th [Diagnosis, Epidemiology, Therapy]	10929
2	(autism spectrum disorder* or ASD or autism).ti,kw.	23890
3	(asperger* syndrome or asperger*).ti,kw.	1150
4	1 or 2 or 3	27668
5	adolescent/ or child/ or child, preschool/	2937612
6	(child or children* or pre-school child* or adolescent*).kw.	70707
7	5 or 6	2940479
8	4 and 7	19625
9	Community Mental Health Services/	18302
10	("child and adolescent mental health service*" or "child & adolescent mental health service*" or CAMHS).ti,ab,kw.	491
11	("child and adolescent mental health team*" or child mental health service*).ti,ab.	149
12	child development clinic*.ti,ab.	39
13	9 or 10 or 11 or 12	18873
14	8 and 13	69
15	Diagnostic Services/	1916
16	(diagnostic service model* or diagnostic assessment model* or diagnostic assessment or diagnostic process).ti,ab.	6616
17	(diagnostic pathway* or diagnostic evaluation or referral pathway*).ti,ab.	8902
18	early diagnosis/ or early intervention/	28062
19	"Referral and Consultation"/	64435
20	Critical Pathways/	6455

21	((multidisciplinary or multi-disciplinary or interprofessional or inter-professional or intraprofessional or intra-professional or interdisciplinary or inter-disciplinary) adj team*).ti,ab.	18640
22	"delivery of health care, integrated"/ or health services accessibility/ or patient care team/	143724
23	Professional-Family Relations/	14480
24	(service delivery or diagnostic experience*).ti,ab.	10451
25	15 or 16 or 17 or 18 or 19 or 20 or 21 or 22 or 23 or 24	283159
26	8 and 25	1254
27	(cost-effectiveness or evaluation).ti,ab.	1027879
28	Efficiency, Organizational/ or Efficiency/	34521
29	evaluation studies as topic/ or program evaluation/ or validation studies as topic/	183858
30	"quality of health care"/ or "outcome and process assessment (health care)"/	95758
31	Waiting Lists/ or Time Factors/	1175790
32	(family experience or parent experience).ti,ab.	365
33	27 or 28 or 29 or 30 or 31 or 32	2377747
34	14 or 26	1295
35	limit 34 to (english language and yr="2011 - 2020")	738
36	33 and 34	256
37	limit 36 to (english language and yr="2011 - 2020")	141
38	remove duplicates from 35	**736**

Appendix B. Data extraction form: papers contributed to PTs

- Appendix B1 - Key papers from primary search and background search
- Appendix B2 - Key papers from secondary searches