DIAGNOSIS OF CO-OCCURRING AUTISM AND ATTENTION DEFICIT HYPERACTIVITY DISORDER

BY

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ABSTRACT

Autism and attention deficit hyperactivity disorder (ADHD) both have onset in early childhood and have multifaceted lifelong effects. The two conditions are separated by different core behavioural symptoms and clinical management. However, autism and ADHD have significant co-occurrence rates. Both autism and ADHD are associated with social differences, high sensory needs, attention difficulties and increased co-occurrence with other conditions. This has resulted in debate about whether autism and ADHD should be viewed as one condition on a gradient or as two separate conditions. Regardless of such the taxonomic classification issues, it appears that, in those children most affected by autism + ADHD characteristics, there are additive challenges across the behavioural and cognitive domains associated with both conditions.

Autistic and ADHD children both benefit from early support, which can reduce some of the negative outcomes associated with these conditions. However, the opportunities to access early support during a sensitive developmental period are reduced by the delayed diagnosis of autism that happens when ADHD co-occurs. A literature review conducted as part of this thesis established that this delay is observed across international jurisdictions. The literature review also demonstrated that there is evidence for an earlier ADHD diagnosis when autism is present. There was also some limited evidence for later age of parental first concern and longer wait time to diagnosis for the autism + ADHD group. Other characteristics which further exacerbated the delay were being female and an ADHD diagnosis being given before the autism diagnosis.

The literature review indicated an international pattern in delayed diagnosis of autism for autistic + ADHD children and that there did not appear to be any studies examining the diagnostic pathway for these children in New Zealand. Therefore, understanding whether this delay is relevant for autistic + ADHD children in New Zealand is an important area of study.

As is a further understanding of the mechanism by which autism + ADHD children might present differently to children with a singular diagnosis of either autism or ADHD. Variables indicated by the literature review, such as wait time to diagnosis and age of parental first concern, were also explored as possibly contributing to delay in autism diagnosis when ADHD was present. In the first study of this thesis, 288 New Zealand parents of children diagnosed with autism (n = 111), ADHD (n = 93), or both conditions (n = 84) participated in an online survey. The results indicated that co-occurring autism and ADHD was associated with an earlier ADHD diagnosis and a later autism diagnosis in New Zealand, thus fitting the international trend. Parents of children with both diagnoses reported less atypical development in language and social behaviours compared to parents of autistic children. The co-occurring group also experienced longer times to diagnosis, and saw more types of specialists prior to a diagnosis than those with autism.

The experience of parents might also enhance an understanding of delays and challenges in the diagnostic process. Therefore, the second study of this thesis used qualitative reflexive-thematic analysis to understand the experiences of parents on their journey to diagnosis for their children. The findings suggest that parents view the process to diagnosis as an adversarial one, which required extensive knowledge, finances, and network resources. Parents also described a second ‘battle’, which was often unexpected, to get support for their child post-diagnosis. Parents identified that the relief of the removal of parenting guilt and blame, as well as situating the issue outside of the child, was an important part of the diagnosis label.

The literature review and both studies one and two indicated that the diagnostic process for autism + ADHD is complex. In the final study, these complexities were further explored by an anonymous online survey of clinicians' (psychologists, paediatricians, psychiatrists) responses (n = 29) to two complex neurodevelopmental cases described in

vignettes. These vignette cases included autism + ADHD characteristics and a number of other co-occurring characteristics. Clinicians reported multiple possible lines of inquiry including specific assessments for particular hypothesised diagnoses and more general assessments. The results of this study suggested that complex cases require lengthy consideration and multiple resources.

The thesis identified and elucidated the phenomena in the timing of diagnosis when autism and ADHD co-occur. This included understanding the delay in autism diagnosis when ADHD is present and provided possible evidence for where along a diagnostic pathway such a delay might be occurring. A biopsychosocial framework was also used to consider multiple psychological and social aspects of this process including the experiences of complex neurodevelopmental cases for families and clinicians. The experience for families and clinicians grappling with an imperfect diagnostic model suggests that multiple resources, time delays and stressful situations are common. This thesis concludes with theoretical and practical recommendations for an improved diagnostic pathway and experience for clinicians and families.

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PUBLICATIONS ARISING FROM THIS THESIS

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DECLARATION BY THE AUTHOR

This thesis is composed of original work in fulfilment of a PhD under the supervision of Dr. Hannah Waddington (primary supervisor) and Dr. Kelly Carrasco (secondary supervisor) from the School of Education at Victoria University of Wellington and Prof. Andrew Whitehouse (external supervisor) from Telethon Kids Institute, Perth Australia. No part of this thesis has been previously submitted for any other degree or diploma. Work by other authors has been referenced in the text and others' contributions have been clearly stated.

Dr. Hannah Waddington, provided input into the design, analysis and interpretation of results and editing of all chapters in this thesis. Dr. Kelly Carrasco and Prof. Whitehouse provided input into parts of the design and editing for each of the studies. Aside from this input, the studies presented in this thesis were designed, analysed and conducted by me.

This thesis includes four original articles that were designed, and analysed by me as part of my thesis by publication. Two articles have been published and one article is in press and the final article is under review. The respective publishing agreements are included as Appendix A in this thesis.

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PREFACE

Overview of Thesis

This thesis has seven chapters and was conceived as a thesis-by-publication. The aim was to publish the literature review and three articles pertaining to the research I completed.

The first chapter is an introduction to co-occurrence before outlining autism spectrum disorder (autism) and attention deficit hyperactivity disorder (ADHD). Each condition is introduced separately before outlining the implications of autism and ADHD co-occurring together (autism + ADHD) and the difficulties of a delayed autism diagnosis when ADHD co-occurs.

The second chapter is a published literature review charting the current information on the delayed diagnosis of autism when ADHD co-occurs. The literature review also examines the research aimed at understanding why this delay of an autism diagnosis might occur. The review also introduces the evidence for an earlier ADHD diagnosis when autism co-occurs.

The third chapter is an overview of the research studies in this thesis including the research questions and methodology.

The fourth chapter is a published quantitative study on parent reported atypical development and age of diagnosis across three groups; autism only, ADHD only and a co-occurring group in New Zealand.

The fifth chapter is qualitative research on parents' experience of the diagnostic process in New Zealand across the three diagnostic groups.

The sixth chapter is research that aimed to understand some of the challenges clinicians' face with diagnosing complex neurodevelopmental cases, as seen with autistic + ADHD children.

The seventh chapter is a general discussion, which summarises the above studies and discusses overarching limitations, implications and suggestions for future research.

I have tried to follow the Autism New Zealand Terminology Resource (Monk, 2022). This includes using the term autism, autistic person, condition, and co-occurrence as opposed to ASD, person with autism, disorder and co-morbidity. I have been on my own journey of learning from autistic people and some of my earlier publications did not adhere to this guidance. Where possible I have changed my terminology use to be in line with their guidance.

CHAPTER 1

INTRODUCTION

Theory of Co-occurrence

Co-occurrence is defined as two or more diagnosed conditions in an individual (Valderas et al., 2009). Researchers differ in the types of diseases, disorders, illness and/or health problems they define as co-occurring, such as whether difficulties with sleeping or eating are included. In their review of co-occurrence in child and adolescent psychiatric conditions, Angold et al. (1999) concluded that increased interest in co-occurrence is connected to the inception of the Diagnostic and Statistical Manual (DSM) and the International Classification of Diseases (ICD), which aimed to reduce complex collections of characteristics into single diagnoses. Particularly in early editions of the DSM, diagnostic hierarchies existed which applied criteria so that co-occurrence was generally precluded (Beauchaine & McNulty, 2013). This led to an obscuring of aetiology, particularly patterns in families, and an oversimplification of symptoms (Beauchaine & McNulty, 2013). Research during the 1980-1990s drew attention to these flaws in the diagnostic hierarchy so that by the third edition of the DSM, most exclusions restricting co-occurring diagnosis were removed (Beauchaine & McNulty, 2013). A notable exception was the co-occurrence of ADHD and autism, which had a diagnostic hierarchy in place until the 5th edition of the DSM (2013), stating that only an autism diagnosis could be given when characteristics of autism and ADHD were present.

One of the impetuses for the creation of the DSM and ICD was to bring international diagnostic consensus to psychology and psychiatry, as prior to this different conditions were defined differently across locations, hampering opportunities for comparative research (Scott & Henry, 2017). The reliability of diagnosis in psychology has significantly improved since the inception of internationally recognised Diagnostic and Statistical Manual (Scott & Henry,

2017). However, the diagnoses do lack predictive validity due to the diagnostic criteria being described using established clinical samples rather than understanding how conditions might develop and change over time in a general population (Scott & Henry, 2017). In some cases this has led to individuals receiving multiple diagnoses over a life-time that might best have been explained by a single diagnosis (Scott & Henry, 2017).

Related to this is the concept of "artifactual co-morbidity," which was a term used in the DSM IV, where there is not true co-occurrence, but rather co-occurrence happens as a by-product of a Diagnostic and Statistical Manual that identifies categorical diagnoses, and does not account for progression of a condition. First (2005) writes, "It is important to understand that comorbidity in psychiatry does not imply the presence of multiple diseases or dysfunctions but rather reflects our current inability to apply Occam's razor (i.e., a single diagnosis to account for all symptoms)" (p. 206). In the USA, the concerns about increases in co-occurrence in the DSM III led to a congressionally mandated National Comorbidity Survey (NCS) conducted between 1990-1992 (Gadermann et al., 2012). In the group with a DSM III condition, over half of respondents had a second condition and a third had three or more conditions. This survey continues to be repeated with increasing rates of co-occurrence recorded (Gadermann et al., 2012).

The study of co-occurrence draws on a number of theoretical models. The first model is the biomedical model, which proposes that all mental disorders are biologically-based brain diseases, which can be thus defined using a classification system such as the DSM (Deacon, 2013). The word "co-morbid" comes from this model, and therefore, will subsequently be referred to as "co-occurrence." The use of the biomedical model within psychology is problematic and heavily criticised (Angold et al., 1999; Deacon, 2013; Follette, & Houts, 1996; Kaplan et al., 2001). It is problematic due to the difficulties with applying the objectivity to observed 'symptoms' from internal mental functioning and then fitting these

'symptoms' into 'disorder' categories. The bio-medical model generally purports that there is an objective test or assessment that allows for the presence or absence of a diagnosis, but diagnosis in psychology appears to be more complicated, often relying on reports of other stakeholders (Scott & Henry, 2017). The biomedical psychology diagnostic pathway is usually focused on the identification of individual differences in a child, a referral to a specialist, an assessment and identification of a DSM 5 label, followed by an evidenced-based intervention provided to the child and possibly the family (Rutherford et al., 2021). The diagnosis is based on clinical judgement and the recognition of a series of symptoms patterns that are conclusive enough for a yes/no judgement (Scott & Henry, 2017).

There are a number of elements that remove the more objective present/absent dichotomy of biomedicine in psychology, such as the fact that the child can have a developmental progression of conditions, show masking of symptoms, not meet thresholds for treatment, and/or have multiple additional symptoms, sometimes called "dimensional symptoms," which further complicate an identification of a "single" condition (Scott & Henry, 2017). In addition, different DSM defined disorders can share common aetiological, behavioural and cognitive features which can make diagnostic identification more problematic (Fusar-Poli et al., 2019). Furthermore, evidence-based treatments and supports often rely on specific isolated DSM disorder categories (Fusar-Poli et al., 2019). This results in treatments and supports siloed with their matched label category rather than applied across relevant conditions (Fusar-Poli et al., 2019). Sometimes the co-occurring condition can also mean that the person is excluded from accessing support, because although the support is applicable to one of their conditions, the criteria for access is that the other condition is not present (Fusar-Poli et al., 2019).

A contrasting criticism of the DSM-5 and ICD-11 is that the system is not biomedical enough, but rather the diagnoses are developed through a collaborative unscientific consensus

of practitioners, which is culturally and socially bound to the time (Frances & Widiger, 2012). Therefore, alternate models for diagnosis have attempted to apply a more scientifically rigorous approach such as the Research Domain of Criteria initiative (RDoC) or a clinical staging model (developmental spectrum model). Both these models have links back to the field of developmental psychopathology, which particularly emphasised the complexity of developmental progression of patterns of "maladaptive behaviour" (Achenbach, 1974) and models in medicine.

The RDoC was developed by the US National Institute of Mental Health (NIMH) with the aim to produce a "biologically based" definition of conditions rather than the symptoms based definitions used in the Diagnostic and Statistical Manuals (Nierenberg, 2019). This new taxonomy for mental conditions has been particularly advanced for schizophrenia, whereby bio-behavioural vulnerabilities are identified that cut across diagnostic-manual definitions (Lindenmayer, 2018). The RDoC has identified five key behavioural domains (negative valence, positive valence, cognitive, social, and arousal/regulatory) which help identify possible "psychopathology" and were considered more informative than diagnosed individual conditions (Beauchaine & Cicchetti, 2016). These domains are linked neuro-biological cortical and sub-cortical emotion and behavioural systems (Beauchaine & Cicchetti, 2016). The discipline of developmental psychopathology and RDoC led to ontogenesis process models, which tried to map complex interactions over time to show the development of more general "externalising behaviours" (Beauchaine & McNulty, 2013). However, it has been questioned whether any progress has been made in explaining complexity or in creating a viable replacement for the diagnostic manual (Nierenberg, 2019).

The clinical staging model (CSM) helps to account for a developmental progression of conditions and reduces the need for multiple co-occurring diagnoses (Scott & Henry,

2017). Multiple conditions have similar onset symptoms, such as initial presentation of attentional difficulties and anxiety. An example might be that a young person, who was initially given an attention and anxiety diagnosis, subsequently goes on to develop bi-polar condition. Although, this would be considered a relatively common onset symptom pattern, the young person might maintain an attention and anxiety diagnosis. Therefore the criticism of the DSM 5 classification system from a CSM position is that it does not account for this level of progression, guide treatment or predict outcomes. The CSM model places pre-eminence on risk factors with an emphasis on evidence-based interventions. This is done by using a clinical staging model from biomedicine that purports there are discrete points in the course of a disease, which reflect severity, impact outcomes, and require different interventions (Scott & Henry, 2017). Staging models have been identified for a number of conditions such as eating, anxiety, and depressive disorders. The clinical staging model has also been used to suggest that rather than individual diagnoses, the field should consider a single staging model for all affective and psychotic conditions because there are factors across all conditions that appear to predict progression (Scott & Henry, 2017). This is seen as a transdiagnostic clinical staging model, and not all proponents of the CSM support this direction (Scott & Henry, 2017).

The RDoC and the clinical staging model attempt to resolve co-occurrence and the flaws of the diagnostic manuals, by adhering more to a bio-medical paradigm. Two further alternative models, transdiagnostic and HiTOP model, also try to resolve these issues, but these models move further away from a bio-medical paradigm. Instead the transdiagnostic and Hierarchical Taxonomy of Psychopathology (HiTOP) models take a more structural or umbrella approach to diagnosis.

The term "transdiagnostic," has been used as an alternative solution to diagnostic manuals with the aim of providing better classification and treatment for mental conditions.

The area has been hampered by a lack of clarity around what constitutes "transdiagnosis" with "trans" meaning "across" diagnosis (Fusar-Poli et al., 2019). Co-occurrence is again a driving force for the inception of the transdiagnostic paradigm. Transdiagnosis, however, came out of the successful use of interventions across diagnoses, in particular cognitive behavioural therapy (Fusar-Poli et al., 2019). A systematic literature review of transdiagnostic research aimed to examine whether there was consistency across the studies to build a viable alternative to the current DSM 5 classification system and found only 3 of the 111 articles met the strict criteria of being "transdiagnostic" (Fusar-Poli et al., 2019). The review was critical for identifying conceptual weaknesses, including that the DSM IV never claimed to create discrete entities and that the distinct boundaries of symptoms have sometimes been simplified and demarcated by others rather than in the diagnosis manuals. In addition, there is the philosophical position that "it would make no sense to challenge the diagnostic boundaries without assuming that these do exist on some ontological level" (Fusar-Poli et al., 2019 p. 202). Although consensus around transdiagnosis is lacking, it is repeatedly referred to in the literature on co-occurrence as solving the future possibility of the classification of conditions.

In 2018, the Hierarchical Taxonomy of Psychopathology (HiTOP) Consortium was established to investigate general dimensions which might sit above individual labels and which might provide better diagnosis and intervention (Krueger et al., 2018). The group defined the diagnostic manuals as authoritative discrete nosology in contrast to their empirical continuous identification (Krueger et al., 2018). This approach has also not escaped criticism. The first outlined criticism is the idea that HiTOP appears to work from the premise that co-occurrence is a problem that needs fixing and that narrowing to one condition might be misleading. A more valid aim of HiTOP might be to hope to reduce 'artifactual comorbidity' (Wittchen & Beesdo-Baum, 2018). The further two criticisms are that

traditional assessment measures, which are a snapshot in time and often use DSM criteria, are used to identify the dimensions of the HiTOP model, and that there is a lack of biological-causal pathways to explain the suggested "higher dimensions" (Wittchen & Beesdo-Baum, 2018).

Biopsychosocial Model

All these models have in common an identification of the lack of validity and the high rates of co-occurrence in the diagnostic manuals. The aims are to provide better diagnoses to inform interventions, yet all of the models have suffered criticism and so far, have not taken hold with any form of consensus that looks likely to replace the diagnostic manuals of the DSM or ICD. A biopsychosocial model does not provide an answer, but suggests that understanding diagnosis requires, not only considering factors of human development, but psychological and social factors (Engel, 1981). Therefore, the biopsychosocial model provides the theoretical underpinning for conceptualising co-occurrence in this thesis. In this model, co-occurrence is conceptualised as multiple diagnoses, but also as the experience of co-occurrence by children, parents, teachers and the community. In addition, a further psychological underpinning is the role of identity and diagnosis of neurodevelopmental conditions, and how this might be complicated by co-occurrence.

The biopsychosocial model postulates that to understand a condition, it must be understood on multiple levels from “molecular to societal” (Borrell-Carrió et al., 2004). The biological, psychological and social aspects of understanding are all of equal importance for conceptualising co-occurrence. Valderas et al. (2009) adheres to a biopsychosocial model and defines axes by which this co-occurrence might be framed: these axes include the nature of the condition, the relative importance of the conditions, the chronology of presentation, and social and economic factors for patients (Valderas et al., 2005).

The nature of the condition might include other health or psychological co-occurrences that might reflect different classification systems (Valderas et al., 2005). In the “relative importance of the conditions” axis, the concept of "index" is questioned, in that reasons for prioritising one condition over the other can be multiple and somewhat arbitrary to the person with co-occurring conditions. In the chronology of presentation axis, models of time and sequence are presented for different ways that conditions can occur throughout development or over a trajectory (Valderas et al., 2009). The effect of the condition trajectories in terms of diagnosis, treatment and prognosis and complex etiological interaction models are also considered.

Patient complexity includes factors such as access to health-care, understanding social and economic factors, and how many acute/chronic conditions are present (multimorbidity/ multi-occurrence). In addition, for children with co-occurring diagnoses, it means understanding both the child’s and the parent’s experience of a co-occurrence diagnosis, such as: which symptoms are, or were, most salient; which ones are, or were, noticed first; and whether co-occurrence now, or at any time, led to any restriction or increase in their access to resources (Valderas et al., 2005). This ties in with an integral part of the biopsychosocial model, that is the perspectives and experiences of people and the influences of the broader environment.

Co-occurrence and Self-Concept

Co-occurrence also destabilises another important trend in the literature which is motivated by people situating diagnoses at the centre of their identity. The biomedical model has described diagnosis as a "biographical disruption" (Bury, 1982), however more recently this has been described in reference to neurodevelopmental conditions as "biographical illumination" meaning an enlightening through identity, personal expectations and social relationships in belonging to a community with others who have the same condition (Tan,

2018). The distinction is made that differences in brain development are closely linked to personhood and are thus not considered separable from the person in the same way as a pathological invasion to a body (Tan, 2018).

The concept of biographical illumination is more radical than how changes in identity after neurodevelopmental diagnoses have usually been framed. The description of identity has more commonly been conceived as a dichotomy of both positive and negative factors post diagnosis (O'Connor et al., 2018; Rasmussen et al., 2020). The identity of the parent of a child diagnosed with a neurodevelopmental condition has also been explored in dichotomous terms as a disruption or "cohesion," which is achieved through an acceptance or understanding of the child's past, present and future as a diagnosed child (Rasmussen et al., 2020).

A systematic review examined qualitative analyses looking at changes to young person's self-concept (beliefs about self) and social identity (beliefs about self from others) after neurodevelopmental diagnosis (O'Connor et al., 2018). A dichotomy is again presented both in the literature review and in the results between benefits and risks of self-concept and social identity post diagnosis. In terms of benefits, self-concept is described as greater understanding and reflection of behaviour that can lead to greater self-management. There was also a benefit of self-legitimisation which was described as situating the condition as something to be taken seriously that absolves blame as "a real disease" (O'Connor et al., 2018). Social identity benefits included a sense of community of others with similar diagnoses, better relationships with others, and a social comparison with others who have "got it really bad" (O'Connor et al., 2020). The risks include a risk to sense of self and a "self de-evaluation" that resulted in reduced self-esteem. Social identity risks included a sense of difference from others, lack of awareness of conditions from others and social stigma (O'Connor et al., 2018).

The systematic literature review takes a transdiagnostic approach across psychiatric diagnoses to examine the effects on self-concept and social identity (O'Connor et al., 2018). It is transdiagnostic because the effects on self-concept and social identity of any psychiatric diagnoses are evaluated together across the negative and benefit categories. However, it is noticeable that some diagnoses, such as autism, are mentioned more frequently in terms of the benefits to self-concept and other diagnoses, such as attention deficit hyperactivity disorder (ADHD), are mentioned more in terms of negatives, such as being more stigmatised than depression and other psychiatric diagnoses (O'Connor et al., 2018). There is no consensus across diagnosed communities as to the approach in examining identity post diagnosis. The distinction has been made that a *condition*-person, such as an autistic person rather than a person with autism, indicates that the condition is socially constructed rather than sits outside the person as an objective entity (Krcek, 2013) or that such framing is an embracing that the condition is not separate but rather something apparent at the core of their being (Tan, 2018).

An important term coined in the early 1990s is that of neurodiversity, which is used to indicate that differences in brain function should be viewed not as impairment, but diversity in the same way that race and ethnicity are examined and valued (Krcek, 2013). Another important part of diversity is acknowledgment that difference, in this case neurological difference, can lead to a majority-neurotypical prejudice (Krcek, 2013). This is a prejudice for 'normal' being the right way to think as opposed to seeing value and understanding of a different way of thinking and understanding the world. However, there is limited research in how co-occurrence might disrupt the dialogue around neurodiversity, biographical illuminations, sense of community, or dichotomous descriptions. As the discussion takes place around identity and diagnosis, co-occurring diagnosis questions demarcations of understanding.

Studying co-occurrence draws on a number of issues, such as the imperfection of a biomedical model, the social experiences of family with co-occurring diagnosis, the sense of biographical illumination or disruption, and an examination of the different developmental trajectories that might be present when a child has a dual or multiple diagnoses. A biopsychosocial model is similar to ecological systems theory in terms of emphasising the importance of social and cultural influences in order for understanding a phenomenon or human development (Bronfenbrenner, 1992), however a co-occurrence diagnosis disrupts or changes the child's experience at all levels from individual to environmental and societal. A common example of a co-occurring diagnosis is that of autism and ADHD. This diagnosis appears to present with the compounding support needs of both conditions. Therefore, this thesis is focused on the complexity of presentation of co-occurring autism + ADHD and the high needs and unique experiences of this population. Each condition will be introduced separately outlining prevalence, diagnosis, co-occurrence, and outcomes, before examining the implications when the two conditions co-occur.

Autism

Definition

Autism is characterised by the DSM 5 as rigidity of behaviour and social deficits, and is considered to be a neurodevelopmental condition (American Psychiatric Association, 2013). Neurodevelopmental conditions have lifelong effects, and usually develop in early childhood. Infantile autism first appeared in the third edition of the DSM in 1980, while the fourth edition named autism as one of five pervasive developmental conditions (American Psychiatric Association, 2000). Of these five pervasive developmental conditions, Asperger's disorder, which was described as a higher functioning autism, and pervasive developmental disorder - not otherwise specified (PDD-NOS), were subsumed under the category of autism spectrum disorder (autism) in the 5th edition of the DSM (American Psychiatric Association,

2013). Thus autism was described as a spectrum or dimensional diagnosis with a range in functioning and severity, rather than pertaining to separate categories for severity between autism and Asperger's disorder (Kuriakose & Shalev, 2016).

Within the two essential domains for diagnosis of autism, rigidity of behaviour and social deficits, the DSM 5 creates several further descriptive criteria (American Psychiatric Association, 2013). In the rigidity of behaviour domain, the DSM 5 describes hyper or hypo reactions to sensory stimulation; an insistence on routine, ritual or sameness; highly restricted interests or obsessions; and stereotyped or repetitive behaviour (American Psychiatric Association, 2013). In social deficits, the DSM 5 describes how deficits can be present in communication and interaction and must be present across multiple settings. Social deficits can include difficulties understanding and displaying non-verbal communication (eye contact, body language), developing and maintaining social relationships, and understanding social-reciprocity (American Psychiatric Association, 2013). Additional notes on the diagnosis of autism in the DSM 5 mention the level of support that a child might need ranging from 'support' to 'substantial level of support' with regards to functioning within a domain. Weitlauf et al. (2014) explore the idea that this is a measure of severity with challenges for consistent application across clinicians. Other notes in the DSM 5 specify the presence or absence of genetic abnormalities, intellectual disability (ID), language impairment, and catatonia (American Psychiatric Association, 2013).

A more strengths based approach to defining autism might also consider the personal strengths of autistic people, which includes skills that are stronger than other abilities for the individual, but not necessarily above a general population (Bal et al., 2022). A focus on personal strengths can lead to better self-esteem and career outcomes (Bal et al., 2022). In addition, autism has also been associated with extraordinary talents (ET) (van't Hof et al.), which are defined as talents that exceed the average abilities of the general population (Bal et

al., 2022). ETs have also been called "savants", "splinter skills" and special talents. ETs remain relatively understudied in an autistic population (Bal et al., 2022). A recent study of 1470 autistic children with IQ scores over 70 found that 46 % of children had a parent reported ET and an a further 23% had a personal strength in the absence of an ET (Bal et al., 2022). Cognitive testing corroborated that these children showed relative strengths in the parent indicated area (visual-spatial, drawing, reading, computation, memory and music) and that some parents even underestimated their child's skill level (Bal et al., 2022).

It is argued that greater acknowledgement of ETs and person strengths might have positive long-term implications for the autistic person (Bal et al., 2022). The ET literature has also been criticised for using arbitrary cognitive testing to verify talents, and that more personal strengths and ET's would be identified if a real-world context was applied (Russell et al., 2019). A qualitative study asked autistic people about their "autistic advantage," with almost all describing some advantageous characteristic, particularly attention to detail, good memory and creativity (Russell et al., 2019). However, the results emphasised that the advantage was context dependent and what could be advantageous could be a negative in a different setting.

Prevalence

A recent systematic review of global autism prevalence states approximately 1 in 100 children are diagnosed with autism in the world (Zeidan et al., 2022). In the USA, the prevalence rate is given by the Centre for Disease Control and Prevention as 1 in 59 children (1.7%) (Rosales et al., 2019). A study of rates of incidence in a particular area of New Zealand (Drysdale & van der Meer, 2020) and a national meta-data study found New Zealand rates comparable to USA data (Bowden et al., 2020).

There is also a higher prevalence of male children diagnosed compared to female children. A meta-analysis suggests that there are three males to every one female diagnosed

with autism (Loomes et al., 2017). There has been speculation about the possible causes of this gender discrepancy (Hull et al., 2020). One possible hypothesis states that females are less likely to develop autism attributed to a female protective effect theory (FPE theory). Another hypothesis is that there is diagnostic identifiers and variation in the way females present (female autism phenotype), meaning there is a diagnostic bias towards males (Hull et al., 2020). The difficulty with FPE theory is that no conclusive evidence has been provided and it is based off the assumption that current diagnosis rates are correct (Hull et al., 2020). There is increasing evidence that female autism characteristics might be different from male characteristics. In terms of core autism characteristics, autistic females may have more challenges in relation to other females, but might show greater social motivation and less social difficulties than autistic males (Sedgewick et al., 2018). Areas of restrictive interest are also thought to be different for females and not as clearly atypical as defined by criteria (Antezana et al., 2019). A final characteristic of female autism phenotype is a greater ability than males to camouflage, which is achieved through masking autistic characteristics using different strategies either intentionally or unintentionally (Hull et al., 2020). As the diagnostic criteria for autism has been developed for a male presentation, recent efforts have focused on trying to account for camouflaging (Tubío-Fungueiriño et al., 2021). Therefore, the evidence suggests that the gender imbalance might be even greater than the current prevalence data suggests.

Autism Diagnosis

Although there can be some variation in Western countries, the common pathway towards an autism diagnosis usually involves parental concerns, recognition by a professional, and diagnostic assessment followed by diagnostic feedback and support (Abrahamson et al., 2021). Research suggests that children as young as 14 months can be reliably diagnosed with autism (Pierce et al., 2019) . However, most autistic children are

diagnosed at around 4 - 7 years of age (Baio et al., 2018; Hess & Landa, 2012; Rosenberg et al., 2011). There are a number of reasons for the delay between when a child's autism diagnosis can be reliably made (14 months) and when children typically get diagnosed. Prior to research on the effectiveness of early intervention, there was less imperative to obtain an early diagnosis and often parents and professionals adopted a wait and see model (Bent et al., 2020). However, current research now focuses on reducing the delay in obtaining an autism diagnosis in order to access effective support at a crucial early developmental period (Whitehouse et al., 2021).

In New Zealand the average age of diagnosis is 6.6 years (Eggleston et al., 2019). Eggleston et al. (2019) found that 53.1% of New Zealand parents expressed satisfaction with the process of receiving an autism diagnosis for their child, which was higher than expressed by parents in the United Kingdom (UK). The overall rate of stress during the diagnosis process was approximately 20% lower in New Zealand compared to the UK (Crane et al., 2016). Eggleston et al. (2019) speculate cultural differences and a lower age of diagnosis (6.6 years in NZ; 7.5 years in UK) might contribute to the higher levels of satisfaction in New Zealand. In addition only 13.8% of parents in a New Zealand sample had their first parental concern dismissed in the initial consultation, while it was 30% in a UK sample (Eggleston, et al., 2019).

Initial dismissal of parental early concerns, or premature reassurance, are associated with parents feeling more stress (Ryan & Salisbury, 2012). Dismissal of parents' first concerns is not only associated with increased parental stress, but it can also contribute to delays in diagnosis (Baio et al., 2018). Eggleston et al. (2019) found that the average age that parents had concerns about their child in New Zealand was 3.2 years. This suggests that there is a considerable delay between parents' first concerns and the average age of diagnosis at 6.6 years. This late diagnosis limits the opportunity for a child to have early intervention.

Internationally, although parents' first concerns range widely, most parents recognise some characteristic of atypical development in their autistic child by 12 months (Waddington et al., 2022). Baio et al. (2018) note that 85% of parents had clinically documented concerns when their child was aged 3, but that only 42% of the children had been given an evaluation by that age. Guinchat et al. (2012) report that the earliest concerns expressed by parents of children later diagnosed with autism were often not related to symptoms of autism, and instead commonly included references to lack of attention, hyperactivity, sleep concerns, sensory reactivity, and atypical emotional regulation. Turygin et al. (2014) found multiple first concerns were predictive of a later autism diagnosis. Kozlowski et al. (2011) note that the mean age of children at the time of the first parental expression of concern had dropped over a 20-year period. The authors speculate that parents are more aware of, or sensitive to, developmental concerns than older generations.

Several studies have looked at a range of parental first concerns, and found that parental concerns regarding speech and communication are the strongest predictors of a subsequent autism diagnosis for their child (Matheis et al., 2017). Becerra-Culqui et al. (2018) examined the relationship between the type of parental first-concern (i.e., those behaviours of the child which were specifically mentioned by the parent) and the time of diagnosis, and found that poor eye contact, pointing/gesturing, responding to name and babbling were associated with earlier diagnosis. Matheis et al. (2017) briefly address the finding that parents whose child has a co-occurring medical condition have earlier parental first concerns than do parents of children presenting with autism only. The examples of medical conditions quoted by Matheis et al. (2017) include seizures, premature birth, cerebral palsy and other genetic conditions.

Co-occurrence

The prevalence and pattern of co-occurring conditions with autism is debated. Research has indicated that the incidence of co-occurring symptoms and diagnoses with autism is as high as 95% (Soke et al., 2018). There are factors which influence these high prevalence rates, including the age of the autistic participants. The high prevalence rate recorded by Soke et al. (2018) increased from 67% co-occurrence for 4-year-olds to 95% for 8-year-olds. In addition, the way that conditions are defined also influences the assessment of their prevalence. For example, Soke et al. (2018) included co-occurring conditions or symptoms such as sleep abnormalities, language disorder, sensory processing, and gastrointestinal problems in addition to genetic conditions, developmental, psychiatric and neurologic diagnoses. Therefore, Soke et al. (2018) state that the high rates of 'co-morbidity' (> 95%) which they recorded might be due to the number and type of co-occurring conditions examined in their study. Simonoff et al. (2008) specified the psychiatric conditions co-occurring with autism as including anxiety, oppositional defiant disorder (ODD), tic disorder, and ADHD, but also included the physical conditions of bowel incontinence (encopresis) and urine incontinence (enuresis) in measurements of co-occurrence. Simonoff et al. (2008) recorded prevalence rates of 70% of autism children having a single co-occurring condition and 41% having two or more.

A challenge for studying co-occurrence in autism is the difficulty of defining what constitutes co-occurrence, isolating the autism phenotype, and the complicating behaviour indicators for autism, such as language disorder and sleep abnormalities. The latter are considered by some to be “soft symptoms” or dimensional characteristics of autism, and by others to be co-occurring conditions (Ramtekkar, 2017). It has also been suggested that there is a false dichotomy between categorical diagnostic criteria and dimensional approaches to autism because the DSM 5 does specify the inclusion of cognitive and language abilities and the recording of co-occurring diagnoses (Rosen et al., 2021). In addition, the categorial

specification of autism might be considered only a starting point for diagnosis, but that dimensions might be emphasised for the autistic person for a particular purpose, such as support services (Rosen et al., 2021).

Outcomes

Studies of lifetime adult outcomes for autistic children, regardless of intellectual ability, show lower rates of achievement in employment, social relationships and independent living than a neurotypical peers (Farley et al., 2018). The majority of autistic adults live with their parents and require increased financial support for this living arrangement after leaving high school (Farley et al, 2018). In this study, only 25–35% of autistic adults were employed full-time (Farley et al, 2018). Farley et al. (2018) describe how autism characteristics tend to decline over time, reaching greatest amelioration at the end of high school, and speculate that this is due to individuals learning more adaptive/camouflage skills throughout childhood. Employment or education is associated with continued reduction in support needs, but many autistic people do not reach their career goals, leading to frustration and increased anxiety and depression in an autistic adult population (Farley et al, 2018). However, with the right employment supports in place, autistic adults can achieve these goals (Brooke et al., 2018). One study showed that if the right supports were in place, such as job customisation, assessment and constructive feedback with opportunities for career advancement, autistic people with moderate to intensive support in their employment moved to minimal support by 18 months of employment (Brooke et al., 2018).

Early intervention is associated with improved levels of functioning, resulting in less support-needs for families and community and better educational and life outcomes (Rosenberg et al., 2011). Early intervention for autism is concerned with accelerating learning in the areas that might be less strong for that child, such as social and communication skills (Rogers and Dawson, 2010). Improvements in these areas can lead to

more general improvements in cognitive functioning so that social learning becomes naturally rewarding (Rosenberg et al., 2011). This has led to a general acceptance of the need for earlier diagnosis, in order for families to access earlier, and therefore more effective, support.

ADHD

Definition

ADHD is defined by a triad of behavioural symptoms: inattention, impulsivity, and hyperactivity which are demonstrated across multiple environments, and which impair the functioning of the child (American Psychiatric Association, 2013). ADHD is also described as a neurodevelopmental condition because it has lifelong effects, usually develops in early childhood, and is multifaceted (Mullin et al., 2013). Underlying the two criteria of inattention and hyperactivity/impulsivity are nine symptoms, each with a descriptive example. Inattention includes difficulty with close attention to details, sustaining attention, listening, following instructions, organising, engaging in effortful tasks, keeping track of belongings, blocking extraneous stimuli, and remembering daily activities (American Psychiatric Association, 2013). Hyperactivity and impulsivity include difficulty staying still, staying seated, running/climbing at appropriate times, engaging quietly in play, resting, talking moderately, being patient, waiting a turn, and resisting intruding or interrupting others (American Psychiatric Association, 2013). The DSM 5 states that the symptoms need to occur often in multiple settings in order to fit the criteria for ADHD (American Psychiatric Association, 2013). It is also specified that these symptoms must be present for 6 months or longer and not due to a "manifestation of oppositional behaviour, defiance, hostility, or failure to understand tasks or instructions" or better explained by another condition (American Psychiatric Association, 2013). There are also three levels of severity; mild,

moderate and severe. The levels of severity are judged by number of symptoms and also the extent of functional impairment for the child.

There are three defined subtypes of ADHD: inattentive (having six or more inattentive ADHD symptoms); hyperactive-impulsive (six or more hyperactive symptoms); and a combined type (six or more of both hyperactive and inattentive symptoms); (American Psychiatric Association, 2013). This is altered to 5 symptoms for an adult to be diagnosed, but the symptoms must have been present prior to age 12 years.

ADHD has also been conceptualised as form of neurodiversity, where viewed deficits are because of a mismatch between the environment and the neurodivergent individual rather than located 'within' the individual (Sonuga-Barke & Thapar, 2021). ADHD has been associated with unique strengths for some individuals such as energy, creativity and entrepreneurial thinking (Sonuga-Barke & Thapar, 2021). There is some evidence for increased elevations of entrepreneurial and creative thinking in an ADHD population compared to neurotypical people (Gonzalez-Carpio et al., 2017; Moore et al., 2021), however the conceptualisation of ADHD as neurodivergent is in its nascence.

Prevalence

The World Federation of ADHD brings together meta data and scientist from around the world to create an international consensus statement on ADHD (Faraone et al., 2021). The most recent statement from the World Federation of ADHD stated current prevalence rates for youth are 5.9% and 2.5% for adults (Faraone et al., 2021). There is also a greater prevalence of males diagnosed with ADHD with two males diagnosed to every one female (Faraone et al., 2021). However, a study across Europe placed ratio rates of girls to boys at much higher levels from 1:3 to 1:16 (Nøvik et al., 2006). Similarly to autism, there is the female protective effect theory that suggests that females require higher environmental and genetic input to reach an ADHD threshold than males (Young et al., 2020). In addition there

is also the theory that females differ from males with less combined type, and more of inattentive presentations of ADHD (Young et al., 2020). Females also show less externalising co-occurring conditions and issues, resulting in seemingly less imperative for others to seek diagnosis (Young et al., 2020). There is some evidence of gender bias in referrals with a study showing that the same vignette of ADHD child with differing genders led to teachers referring the male, but not the female hypothetical student for further support (Sciutto et al., 2004). There is also mention of females masking or camouflaging their ADHD symptoms more successfully than males. Female masking of ADHD is framed as females using compensatory and coping behaviours to 'hide' ADHD characteristics, including socially adaptive behaviour and dysfunctional strategies, such risky behaviour (Young et al., 2020).

Diagnosis and ADHD

Results of a review of diagnostic experiences of ADHD children in the USA indicate that the median age for diagnosis was 7 years, with 1 in 3 children diagnosed before the age of 6 (Visser et al., 2015). No research in New Zealand has reviewed diagnostic experiences of ADHD children or the median age of diagnosis. Internationally, diagnosis before the age of 6 for ADHD is debated because clinical guidelines state that an assessment should be done across multiple settings and school is considered a more appropriate setting for diagnosis than that provided by an early childhood setting (Visser et al., 2015). There are also few diagnostic tools available for use at this early age, with the validity to support a diagnosis (Visser et al., 2015). Most diagnostic tests for ADHD, such as the Conner's Assessment, can be used only from the age of 6 years (Lyall et al., 2017).

This picture is complicated by the use of an effective medication to treat ADHD. Guidelines from the American Academy of Paediatrics and the American Academy of Child and Adolescent Psychiatry for the treatment of younger children recommend that behaviour therapy be the first line of treatment for ADHD rather than medication, but approximately

half of ADHD children in the USA received behaviour therapy while three fourths received medication (Visser, 2016). The common use of medication to treat ADHD has led to a resistance in diagnosing children under 6 years, in view of the limitations of ADHD medication (methylphenidate; Visser, 2016). These include: increased tolerance, the fact that any beneficial effects on the symptoms of ADHD are seen only during treatment, the uncertainty surrounding long-term outcomes of medication, and the common side effects of medication, such as reduced sleep, appetite and growth (Sonuga-Barke et al., 2011). Nonetheless, there is a call for earlier recognition of ADHD symptoms in preschool children in order to access behavioural intervention, rather than medication as a first line of treatment (Sonuga-Barke et al., 2011).

Factors which might contribute to delays in diagnosis have become a focus of recent research. One key factor is the relationship between parents' first having concerns about their child and the age at which their child receives a diagnosis. In a large study of Japanese parents', there was an average time lag of 2.6 years between when parents first had concerns and when a ADHD diagnosis was given (Yamauchi et al., 2015). Interestingly, they hypothesised that more severe symptoms would lead to parents seeking a diagnosis sooner, but they found that more severe behavioural problems were actually associated with a longer time delay to diagnosis (Yamauchi et al., 2015). Ghanizadeh (2007) reported a similar phenomenon and suggested that the parents' reasoning was that the behavioural symptoms were a result of parental or child temperament.

In another study, a vignette of an ADHD combined-type child was presented to parents of children with suspected ADHD and to parents of children with no symptoms of ADHD (Maniadaki et al., 2007). The authors predicted that parents of ADHD-suspected children would rank the hypothetical vignette as impacting on a child's and a family's life more than would be ranked by parents without an ADHD-suspected child, perhaps leading to

a greater tendency towards help-seeking. However, the opposite was true, with parents without ADHD-suspected children reporting that the child in the vignette required more professional support and that the child's behaviour would negatively impact on the family's life and the child's outcomes. This suggests that parents of ADHD children are not necessarily as sensitive to ADHD symptom concerns, which might lead to help-seeking and an early diagnosis, as parents of children who do not display these symptoms.

Co-occurrence with ADHD

Similar to autism, the rates of co-occurrence for ADHD are high, with estimates between 40% and 80% depending on the sample and inclusion criteria for a co-occurring diagnosis (ADHD, 2017). Jensen and Steinhausen (2015) examined a large population (>14,800 participants) of ADHD children and adolescents, with a focus on psychiatric diagnosis: 52% of the sample had at least one co-occurring psychiatric diagnosis, with the most common co-occurring diagnoses being 'disorders' of conduct (16.5%), learning, language and motor development (15.4%), and autism (12.4 %). The causal pathway of these additional diagnoses is not clear: for example, ADHD, (2017) speculate whether: (i) inattention may have resulted in a greater disengagement with learning, leading to higher co-occurrence with learning disabilities; (ii) impulsivity and hyperactivity have resulted in greater defiance behaviour; or (iii) the conditions cluster according to shared environmental and genetic risk factors (ADHD, 2017). Rucklidge et al. (2016) examined psychiatric co-occurrence in an adult ADHD population in New Zealand and found comparable figures with overseas data. In an ADHD adult population, 83% had an additional co-occurring psychiatric diagnosis with the most common being a depressive disorder (Rucklidge et al., 2016).

Outcomes

Children with ADHD have negative outcomes compared to children not diagnosed with ADHD, including educational under-achievement, difficulties with employment, an

increased incidence of addiction, challenges with interpersonal relationships, driving offences, antisocial behaviour, and criminality (Faraone et al., 2021). There are improvements in outcomes when children have received treatment, particularly with regard to relationships, but not with addiction and antisocial behaviour (Shaw et al., 2012). There is some evidence for improvements in educational achievement (performance at school and academic achievement on tests) with treatment (Arnold et al., 2020). Educational achievement, and support for ADHD appears to result in greater positive self-esteem, social function and life-time outcomes (Harpin et al., 2016).

Recent research has begun to look at early support for children with ADHD. There are promising results for the effectiveness of parent coaching, executive function training, and behaviour interventions for helping children identified in preschool as being at risk of developing ADHD (Charach, et al., 2013; Feil et al., 2015; Halperin et al., 2020). Therefore, the promising success of early support has led to conclusions that children, who have a high likelihood of ADHD, should be identified in preschool (Charach, et al., 2013; Feil et al., 2015; Halperin et al., 2020).

Co-occurring Autism and ADHD

Definition

Historically, the Diagnostic and Statistical Manual of Mental Disorders (DSM) excluded a dual diagnosis of autism and ADHD (American Psychiatric Association, 1994). The reason being that the frequency of autism occurring with ADHD characteristics, in both a clinical and general population, meant ADHD characteristics were viewed as part of an autism presentation (Ames & White, 2010; Cooper et al., 2014). Therefore, prior to 2013 and the publication of the 5th edition, the co-occurring diagnosis of ADHD and autism went against the specifications of the DSM. In the DSM IV, hyperactivity, short attention span and impulsivity were listed as “associated features" of autism and ADHD was not to be diagnosed

if the symptoms of inattention and hyperactivity occurred exclusively during the course of a Pervasive Developmental Disorder (American Psychiatric Association, 1994). Kentrou, et al. (2019) attribute the DSM IV restriction on diagnosis of autism + ADHD as the cause for the pithy research in understanding this co-occurrence.

The relative salience of the different conditions is also important for defining their co-occurrence. For example, in examining ADHD and autism, the question arises as to which diagnosis should be considered of primary importance so that the other diagnosis is considered co-occurring. Van de Meer et al. (2012) define participant groups according to the pre-eminence of symptoms, thereby distinguishing between an ADHD(autism) and an autism(ADHD) group, each group being defined by a different primary index; however, in most of the studies reported in the literature, autism is considered the index with ADHD having the secondary status of the additional co-occurring diagnosis.

Co-occurring diagnoses are further complicated when there are supplemental diagnostic subgroups involved. In the literature on autism co-occurrence. the distinction is sometimes made between different ADHD subtypes within trials (e.g., Mayes et al., 2012); at other times, however, this distinction is not made, as in the study by Nydén et al. (2010) on the types of neuropsychological conditions most commonly associated with autism, ADHD and combined autism + ADHD in adults. Nydén et al. (2010) found that reaction time variability (associated with the impulsivity symptom of ADHD), was not seen in the autism + ADHD group, and speculated that this was because inattentive ADHD (not impulsivity/hyperactivity symptoms) was more prominent in the co-occurring group (autism + ADHD) than in a general ADHD population.

The high co-occurrence of autism + ADHD co-occurrence, coupled with the previous prohibition by international classification systems of this dual diagnosis, has led to a split in the literature, with some researchers starting from the premise that autism and ADHD are

separate conditions (Separate Disorders Hypothesis) while others support examining the idea that ADHD characteristics are part of an overarching diagnosis of autism (Gradient Disorder Hypothesis) (van der Meer et al., 2012). The gradient hypothesis is that ADHD and autism are part of the same condition with a gradient from ADHD-only moving to autism + ADHD group (Kushki et al., 2019; Rodriguez-Seijas et al., 2020).

Prevalence

In the gradient hypothesis an ADHD only group exists, because in a population of children with the primary diagnosis of ADHD, only 12–25% of such children show autism characteristics (Antshel et al., 2016; Cooper et al., 2014; Jensen & Steinhausen, 2015; Kotte et al., 2013). This contrasts with a co-occurring prevalence of ADHD characteristics in a autism population of up to 80 % (Ames & White, 2011; Leyfer et al., 2006; van der Meer et al., 2012). These prevalence rates support the gradient hypothesis that while ADHD symptoms can occur without autism, autism is less likely to occur without ADHD characteristics. Johnson et al. (2015) have presented the idea that similarities between ADHD and autism symptoms result from common neurological reactions used to compensate for disturbances in early brain function. In such a view, the similarities seen in the two conditions represent common brain adaptations to different forms of brain dysfunction, rather than indicating that the conditions are the same.

Assessing the prevalence of co-occurring autism + ADHD is also challenging because it is dependent on whether diagnosis is required for inclusion of an individual in a study, or whether a symptom assessment is to be given to participants. In the study of Soke et al. (2018), over a quarter of 8-year-olds who had been diagnosed as autism also had an additional diagnosis, given by a clinician, of ADHD. However, if ADHD symptoms are measured in an autism population, results are as high as 80% of that population fulfilling an additional ADHD diagnosis (Ames and White, 2011; Leyfer et al., 2006; van der Meer et al.,

2012). In New Zealand, a study looking at diagnosis over a three year period in the Hutt Valley found that ADHD was the second most common diagnosed co-occurrence at 21.3%, behind speech language impairment (Drysdale & van der Meer, 2020). Another New Zealand study, which used a number of nation-wide sources to create an integrated data set, placed the prevalence rate for a diagnosis of autism + ADHD within an autistic population at 26.8% (Bowden et al., 2020). In Australia, a study looking at the use of medication in an autism population found that 22% of autistic children also had an ADHD diagnosis (Rasmussen et al., 2019).

The literature review by Mannion and Leader (2013) on autism co-occurrence concluded that the prevalence rates for ADHD in an autism population were between 14% and 78%. More recently the estimate for co-occurrence has been between 40–70% (Antshel et al., 2016). Although ADHD is the most common psychiatric co-occurrence for autistic children (Antshel et al., 2016), the wide range in prevalence estimates for this co-occurrence reflects the inconsistency in the population samples and in the measurement techniques used. In addition, clinicians may have been adhering to a practice that reflects previous DSM IV specifications that the two diagnoses cannot co-occur.

Diagnosis

The presence of co-occurring autism + ADHD appears to influence the age of diagnosis (Jensen & Steinhausen, 2015; Soke et al., 2018). An autism diagnosis appears to occur later when ADHD is present and an ADHD diagnosis earlier when autism is present compared to the age of diagnosis when a child is given one of the conditions (Jensen & Steinhausen, 2015; Soke et al., 2018). It is possible that the co-occurring diagnosis is given at the same time or that autism is diagnosed first, however it appears that it is most common to be given an ADHD diagnosis prior to an autism diagnosis when the two co-occur (Miodovnik et al., 2015). Research suggests that the presence of co-occurring ADHD generally leads to a

later diagnosis compared to developmental regression that leads to an earlier autism diagnosis (Jónsdóttir et al., 2011; Soke et al, 2018). Miodovnik et al. (2015) found that children diagnosed with ADHD waited an average of 3 years for the additional diagnosis of autism to be made, and that this was consistent across childhood and independent of the 'severity' of characteristics.

In a pioneering study, Davidovitch et al. (2015) looked at children who were later diagnosed with autism after an earlier multi-team assessment failed to recognise the condition. They found that almost half the children in the sample had initially been diagnosed with ADHD or as showing ADHD symptoms in the first assessment (Davidovitch et al., 2015). Kentrou et al. (2019) asked a large autistic participant group of 2200+ people in the Netherlands, if they had initially had an ADHD diagnosis. ADHD had initially been diagnosed in 12.1% of children in their cohort with an average delay in autism diagnosis of 1.8 years compared to autistic children without an ADHD diagnosis. They found that this delay was significantly higher for females at 2.6 years.

Kentrou et al. (2019) propose two theories for the delay in autism diagnosis when ADHD is present. The first of these is that ADHD is a misdiagnosis, as they note that 40% of the autism sample, who were initially diagnosed with ADHD did not retain their ADHD diagnosis, whereas the remaining 60% retained both diagnoses:

> Children who later go on to receive a diagnosis of autism are initially diagnosed with conditions such as ADHD not only because symptoms of autism are present but missed, neglected, or misattributed to ADHD but because children may not yet manifest the full spectrum of autism symptoms necessary to qualify for a formal diagnosis. This might make clinicians more inclined to ascribe existing symptoms to conditions other than autism or refrain from diagnosing autism until clear and undisputed symptoms are present. In other words, previously diagnosed

> conditions can mask emerging autism symptomatology, biasing attribution of progressively increasing impairment toward existing diagnoses (Kentrou et al., 2019 p. 1069).

The second hypothesis put forward by Kentrou et al. (2019) in the above excerpt suggests a developmental trajectory for an autism + ADHD group in which the ADHD characteristics are initially more salient whilst the autism characteristics become more prominent later. Miodovnik et al. (2015) adds another hypothesis, speculating that a possible cause of the delayed diagnosis of autism in a autism + ADHD population could be that clinicians may feel more confident in diagnosing ADHD due to it being more common: they see more of ADHD behaviour during clinic visits and are more comfortable diagnosing ADHD rather than autism. The authors explain that the bias to diagnose ADHD "is especially true if they [clinicians] have more experience managing and treating ADHD, if they do not feel comfortable giving a diagnosis of autism, or if they lack access to specialists who can conduct comprehensive neurodevelopmental assessments" (Miodovnik et al., 2015, p. 834). Furthermore, Miodovnik et al. (2015) state that 25% of clinicians in the USA did not use the DSM criteria (American Psychiatric Association, 2013) to diagnose ADHD, and only 67% used standardized rating scales. Miodovnik et al. (2015) point out that ADHD is a difficult diagnosis to make due to there being multiple possible causes of attention difficulties. The diagnosis is also particularly difficult to make in young children due to some characteristics being developmentally appropriate in a younger age group.

In Eggleston et al.'s (2019) study of parents' experiences of their child receiving an autism diagnosis in New Zealand, only ADHD was predictive of significantly less satisfaction with the diagnostic process. They found that with the presence of ADHD, children waited longer on a wait list, saw a higher number of professionals prior to diagnosis, and were less satisfied with post-diagnostic supports (Eggleston et al. 2019). However, this

study did not quantify if there was a delayed diagnosis of autism in an autism + ADHD group compared to an autism only group. In addition, the reasons behind this protracted process, and the lower levels of satisfaction with diagnosis expressed by parents in New Zealand, require further investigation.

Whilst the results from these studies (Davidovitch et al., 2015, Kentrou et al., 2019; Miodovnik et al., 2015) suggest there is a significant delay in a diagnosis, there remains speculation as to why clinicians fail to diagnose autism when ADHD is present. A greater understanding of the deliberative processes of clinicians with regard to the profile of a child who presents with autism + ADHD is warranted. Another possible mechanism of this delayed diagnosis of autism might lie in the nature of a parent's first concern, and specialist seeking behaviour. When and what parents first view as a developmental concern can lead to either early, or late, seeking of professional advice (Waddington et al., 2022).

Outcomes

The evidence suggests that children with a co-occurring diagnosis of autism + ADHD experience poorer educational outcomes than do children with only one of the diagnoses (Rao and Landa, 2014). Rao and Landa (2014) investigated the co-occurrence of ADHD and autism in children aged between 4 and 8 years and found that, compared with autistic children, those with the two conditions displayed lower levels of cognitive functioning, increased social impairment and greater delays in adaptive functioning. The cognitive impairments in executive dysfunction appear to be additive, in that an autistic + ADHD child has executive dysfunction that is characteristic of both condition profiles (Berenguer et al., 2019). Parents report that autistic + ADHD children have worse impairments across all adaptive domains and that the children are more impaired than autism-only or ADHD-only groups (Antshel et al., 2016). However, outcomes for co-occurring autism + ADHD in an educational setting are not well understood. Lyall et al. (2017) noted that:

> Follow-up studies of educational attainment for children with combined autism and ADHD symptoms are needed to determine the impact of such symptoms on academic achievement and other crucial outcomes. That the deficits in cognitive and adaptive behaviours appear in pre-school children is perhaps most concerning (Lyall et al., 2017, p.9).

Some school-based measures have shown that autistic + ADHD children were at greater risk of bullying others (Montes & Halterman, 2007) than was the case for autistic children, and also to be ranked higher by both teachers and parents on scales of oppositional defiance (Guttmann-Steinmetz et al., 2009). Other research involving adolescents has demonstrated that those with autism + ADHD are also more likely to be bullied than either diagnosis alone (Zablotsky et. al., 2013). Whether instigating the bullying or being the victim, these children appear to be more prone to challenges with navigating a social milieu and to show more externalising symptoms.

Over time there is a reduction in conduct problems in autistic children, while the conduct problems seen in an ADHD + autism group not only persist but also increase incrementally (Colombi and Ghaziddin, 2017; Flouri et al., 2015). This is a similar pattern to that seen in sensory processing, whereby autistic children show a reduction over childhood in sensory issues, while autism + ADHD children show an increase over their lifetime in sensory processing issues (Cheung & Siu, 2009). Other studies show a greater incidence of tantrums in this group and also higher externalizing behaviour scores compared to groups with autism only and control groups (Craig et al., 2015).

The presence of ADHD with autism resulted in significantly worse outcomes in a social skills training programme than were seen in participants with just autism (Antshel et al., 2011). The basis of these social issues may be that emotional recognition deficits in an autism + ADHD population appear to be worse than in an ADHD or autism only population

(Oerlemans et al., 2014). Unsurprisingly, families of autism + ADHD children experience a lower quality of family life, and have greater parental stress than is demonstrated by families of children with single autism or ADHD diagnoses (Green et al., 2016).

Research has shown that co-occurring ADHD heightens autism characteristics (Ames & White, 2011; Holtmann et al., 2007; Lyall et al., 2017), which suggests that the presence of the two co-occurring conditions complicates the overall presentation of the primary index (in this example, autism). Lyall et al. (2017) argue that ADHD characteristics influence other behaviours within autism and speculate about the directionality of effects, suggesting that ADHD characteristics may impact behaviour and cognition, or that greater cognitive and behaviour support needs may lead to more evident ADHD symptoms. Avni et al. (2018) confirmed that in 260 participants with a mean age of 7.5 years, the co-occurrence of ADHD symptoms with autism was associated with greater challenges in socialisation and adaptive skills, and they called for clinicians to diagnose co-occurring conditions with autism early.

Similarly, Lyall et al. (2017), assessed ADHD symptoms in an autism population of 2 to 5 year-olds and concluded that, in autistic children, there was a significant association between the presence of ADHD characteristics and greater cognitive/behavioural challenges. The degree of inattention was significantly associated with greater cognitive and behavioural needs for a young autism population, but the correlation was significantly less strong for hyperactivity/impulsivity. Lyall et al (2017) add that early support for autistic + ADHD children should specifically target increasing attention for this subset of autistic children.

A few studies have examined an ADHD population for autism traits and have found that more autistic traits in an ADHD population was associated with increased oppositional behaviour, anxiety and general motor difficulties, as well as, lower IQ and working memory (Cooper et al., 2014; Green et al., 2016). Similarly, Tureck et al. (2013) found that increased autism traits was a predictor of inattention and impulsivity in toddlers.

In conclusion, children with a co-occurring autism + ADHD diagnosis have significantly more challenges in cognitive, social and adaptive functioning, and autism mannerisms, than if either diagnosis had been made alone (Holtmann, et al., 2007; Lyall et al., 2017; Rao & Landa, 2014; Tureck et al., 2013). This high need population might receive a diagnosis of autism at a much later age than autistic children without ADHD, which reduces the opportunities for early intervention. Like any neuro-divergent population, they can thrive in an educational, social and emotional context with the right individual and environmental evidenced-based supports (Simon, 2016).

A literature review is required to assess the extent of the delay in autism diagnosis when ADHD is present and to quantify if there is possible evidence for contributing factors to this delay. The inverse implication of an early diagnosis for ADHD when autism is present is also not well understood. An understanding of the relationship between diagnosis and co-occurring conditions will elucidate case complexity and help families and clinicians gain access to support more quickly.

CHAPTER 2

A Systematic Literature Review: Age of Diagnosis for Co-occurring Autism and Attention Deficit Hyperactivity Disorder During Childhood and Adolescence

Chapter Note

This section is based on the published literature review in *Review Journal of Autism and Developmental Disorders,* titled, *Age of Diagnosis for Co-occurring Autism and Attention Deficit Hyperactivity Disorder During Childhood and Adolescence: a Systematic Review*. Dr. Hannah Waddington helped with the design, analysis and editing. Lauren McNeil provided the interobserver agreement. Prof. Andrew Whitehouse and Dr. Kelly Carrasco assisted with editing. I designed, conducted and wrote this review and it is entirely my work aside from the input specified above. Aside from changes to ensure consistency of formatting with the rest of the thesis, this chapter is identical to the published article. The complete citation for the published article is: Sainsbury, W.J., Carrasco, K., Whitehouse, A.J.O., McNeil, L., Waddington, H. (2022) Age of Diagnosis for Co-occurring Autism and Attention Deficit Hyperactivity Disorder During Childhood and Adolescence: a Systematic Review. *Review Journal Autism and Developmental Disorders*, 1-13. https://doi.org/10.1007/s40489-022-00309-7

Abstract

Early identification and intervention are recognised as important elements of the clinical pathway for autism. Autistic and attention deficit hyperactivity disorder (ADHD) children may be diagnosed at a different age than children who only have one of these diagnoses. This systematic review aimed to identify the age at which children were diagnosed with both autism and ADHD. Of the 9552 articles screened, 12 were included in the review. The findings suggest that autism is typically diagnosed later when ADHD is present, and ADHD is typically diagnosed earlier when autism is present. Further research is needed to

understand the factors impacting a delayed autism diagnosis and an earlier ADHD diagnosis when the two conditions co-occur.

Keywords

Co-occurring conditions, diagnosis, autism, attention deficit hyperactivity disorder, age of diagnosis.

Autism spectrum disorder (autism) is characterised by impairments in social communication and the presence of fixed, rigid behaviours (American Psychiatric Association, 2013). The number of autistic individuals who also have an attention deficit hyperactivity disorder (ADHD) diagnosis ranges from 59-83% (Joshi et al., 2020).

Evidence suggests that autistic and ADHD children are a high-need population (Hong et al., 2020; Zablotsky et al., 2020). They appear to have poorer outcomes overall and reduced response to social skills intervention than children with either diagnosis alone (Elwin et al., 2020; Fleming et al., 2020; McDougal et al., 2020). Co-occurring ADHD is associated with more severe autism symptoms, lower adaptive functioning and greater cognitive impairment compared to an autism diagnosis alone (Yerys et al., 2019; Zachor & Ben-Itzchak, 2020). While emotional and conduct challenges for autistic children appear to reduce over time, these challenges appear to increase for children with autism + ADHD (Flouri et al., 2015). Co-occurring ADHD also appears to further exacerbate stress, financial and time burdens for families (Dovgan & Mazurek, 2019).

There is preliminary evidence that obtaining an autism diagnosis may be delayed when ADHD is co-occurring (Davidovitch et al., 2015; Frenette et al., 2013; Jónsdóttir et al., 2011; Miodovnik et al., 2015). This delay may have implications for longer-term outcomes, given the known importance of prompt intervention in early childhood (Towle et al., 2020). This delay in diagnosis seems counterintuitive given that outcomes for children with autism and co-occurring ADHD (hereafter referred to as autism + ADHD) are more severe, and

severity typically leads to earlier help-seeking behaviour and, ultimately, diagnoses (Miller et al., 2021). Data provided by the Centre for Disease Control and Prevention (2014), states the average age of diagnosis for autism is 4 years 4 month and the average age for diagnosis of ADHD is 7 years (Visser et al., 2014), which suggests that autism is generally diagnosed prior to ADHD. The DSM 5 specifies that the symptoms must be present in childhood for both autism and ADHD (under the age of 12 years) (Kooij et al., 2019). Late diagnosis of autism is usually defined as occurring during adolescence (Hosozawa et al., 2020). Delayed diagnosis of adults is beyond the scope of this review. The missed diagnosis of an adult group has been described as a "lost generation" and provides a number of challenges including retrospectively collecting a developmental history of symptoms present in early childhood (Lai & Baron-Cohen, 2015). There is also controversy over whether adult onset of ADHD can occur (Mucci et al., 2018).

It has been hypothesized that the presence of ADHD symptoms may make identifying autism behaviours more challenging in the diagnostic process (Soke et al., 2018). Indeed, studies that have examined missed diagnoses of autism suggest that co-occurring ADHD may be a contributing factor (Davidovitch et al., 2015; Fusar-Poli et al., 2020; Hosozawa et al., 2020). In a study of the objectivity of a widely used clinical tool that informs autism diagnosis, Autism Diagnostic Observation Schedule (ADOS), only 25% of children with autism + ADHD were correctly identified using this measure (Zander et al., 2016). Park et al. (2014) found that the distinguishing factor between those referred for autism and diagnosed with autism, and those referred for other reasons, but subsequently diagnosed, was that the former group had higher support needs with significantly lower externalising and hyperactive behaviour. They hypothesise that externalising behaviours and hyperactivity may mask autism symptoms and that externalising and hyperactive behaviours are more likely to occur if the autistic individual has higher adaptive functioning (Park et al., 2014).

The influence of autism on the timing of the ADHD diagnosis, when autism and ADHD are co-occurring, has received less attention in the literature (Miller et al., 2018). There is also less emphasis on the importance of early identification of ADHD. This may be related to multiple factors, such as that (i) ADHD symptoms may be seen as developmentally typical in early childhood, (ii) the impact on functioning may not be evident until the child reaches school, and (iii) assessments for identification require teacher input from school age (Visser et al., 2014) (Silva et al., 2015) (Miller et al., 2018). Nonetheless, there is increasing interest in earlier diagnosis and non-medical intervention for younger children with ADHD (Hatakenaka et al., 2016; Miller et al., 2020).

A systematic literature review was conducted with the aim of elucidating whether there is a significant difference in age of autism diagnosis for children and adolescents (0 - 21 years of age) with autism only and co-occurring autism + ADHD. The review also sought to compare the age of ADHD diagnosis for children and adolescents with autism only and those with co-occurring autism + ADHD. This will allow an examination of the size and variation of any differences. If differences in age of diagnosis are found, this review will also examine quantifiable factors which may contribute to this.

Methods

This systematic review followed the procedures outlined in the Preferred Reporting Items for Systematic Review and Meta-Analyses (PRISMA) statement (Moher et al., 2009)

Protocol

The study protocol was registered on the 31st of December, 2020 (PROSPERO 2020 CRD42020222984). https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42020222984. The protocol was subsequently updated (13th of May, 2021) to state that all included studies must make a comparison between the age of diagnosis or onset between an autism + ADHD group with an autism only, an ADHD only group or a whole sample.

Eligibility

Studies were eligible for inclusion in the systematic review if they met all of the following criteria.

1. They included children and adolescents up to the age of 21. If individuals over the age of 21 were included, individuals who were 21 and under were analysed separately. This was to distinguish between a delay in acquiring an autism diagnosis or ADHD diagnosis, when a child or adolescent has entered a diagnosis pathway from an adult diagnosis where no diagnosis might have been sought during childhood or adolescence or a misdiagnosis might have occurred.
2. There was a defined co-occurring group of individuals who were diagnosed with autism + ADHD, and a group of individuals diagnosed with autism only and/or ADHD only.
3. There was a comparison between the age of diagnosis or onset between an autism + ADHD group with an autism only, an ADHD only group or a whole sample.
4. The study was peer-reviewed written in English, and published after 2014.
5. The methodology used included a comparison of age of diagnosis across more than one participant (e.g., brief report, review, case studies were excluded).

Search Strategy

The literature search was conducted on the 30th of October 2020 using PsycINFO, MEDLINE, Scopus, and Pubmed. The search was limited to peer-reviewed studies published in English after 2014. The search terms were (diagnos* OR misdiagnos*) AND (autis* OR AUTISM* OR “autistic disorder*” OR Asperger* OR "pervasive developmental disorder*” OR PDD-NOS) AND ("Attention Deficit Hyperactivity Disorder" OR "Hyperkinetic Disorder" OR ADHD OR "Attention Deficit Disorder" OR ADD). In each database, the search was limited to titles, abstracts and keywords. A grey literature search was also

conducted during the week of February 15th, 2021 using the Google advanced search engine (limited to first 100 results per search) and abstracts submitted to International Society for Autism Research (INSAR) conferences in the past 5 years. All searches were limited to articles.

Study Selection

One reviewer (WS) conducted all database searches and imported the results into Covidence reference management software, where duplicates were removed. The same reviewer screened all the titles and abstracts of the studies against the inclusion/exclusion criteria and a second reviewer (LM) screened 20% of the total studies against the same criteria. Then the first reviewer screened full-texts of all potentially relevant articles and the second reviewer screened 20%. Following each stage of screening, the reviewers discussed and resolved disagreements. If a consensus could not be reached, an additional reviewer (HW) was consulted. The percentage of agreements was calculated using the formula: agreements/(disagreement + agreements) x 100. Agreement on the title/abstract screen was 98.5% and agreement on the full-text screen was 92.5%

Data extraction

Data from each study were extracted and summarised in three tables in Microsoft® Word. The following data were extracted for eligible original studies: (i) the total number of participants, (ii) the number of participants in each diagnostic group, (iii) participant gender, (iv) geographical location, (v) the date range of the participant data set, (vi) how the diagnosis was reported (diagnostic determination), (vii) description of the participant population (viii) the age range of the participants, (ix) the age of AUTISM and ADHD diagnoses for each diagnostic group, (x) statistical significance of any differences in age of diagnosis, (xi) the age of parental first concern and (xii) diagnostic wait times for each diagnostic group, (xiii) quantitative factors related to age of diagnosis. The data for all studies

were independently extracted by one reviewer (WS). Reviewers (LM & HW) screened 40% of the total studies for extraction data resulting in an agreement rate of 94.1%.

Risk of Bias

The risk of bias of included studies was assessed using the Hoy et al. (2012) guidelines. The indicators of high quality studies included in this review were: (1) target population was representational of a national population; (2) sampling frame was representative of target population; (3) random selection was used to select a sample; (4) non-response bias minimal; (5) data collected directly from subjects; (6) acceptable case-definition of ADHD or autism; (7) same mode of data collection used for all subjects; (8) prevalence period; (9) no errors in reporting. One item "study instruments have reliability and validity" was not included in this study because the primary outcome, age of diagnosis, did not require a validated measure. The item related to prevalence period (9), was modified to reflect the time that the study allowed for a co-occurring diagnosis to occur whereby low risk was considered greater than a 2-year window. A score of "yes"/0 for indicated low risk for that item and "no"/1, indicated high risk for that item. Studies with scores of 0 - 3 were deemed "low risk", those with scores of 4 - 6 were "moderate risk" and those with scores of 7 - 9 were "high risk". If there was insufficient information in the article, but in-depth protocol was referenced elsewhere, such as a National database, then this was sought. When more information was needed, however and this was not clearly stated in the article, by default the indicator was noted as high risk. Two reviewers independently evaluated 20% of the articles for the quality indicators. All discrepancies were discussed and resolved and the overall agreement was 90%.

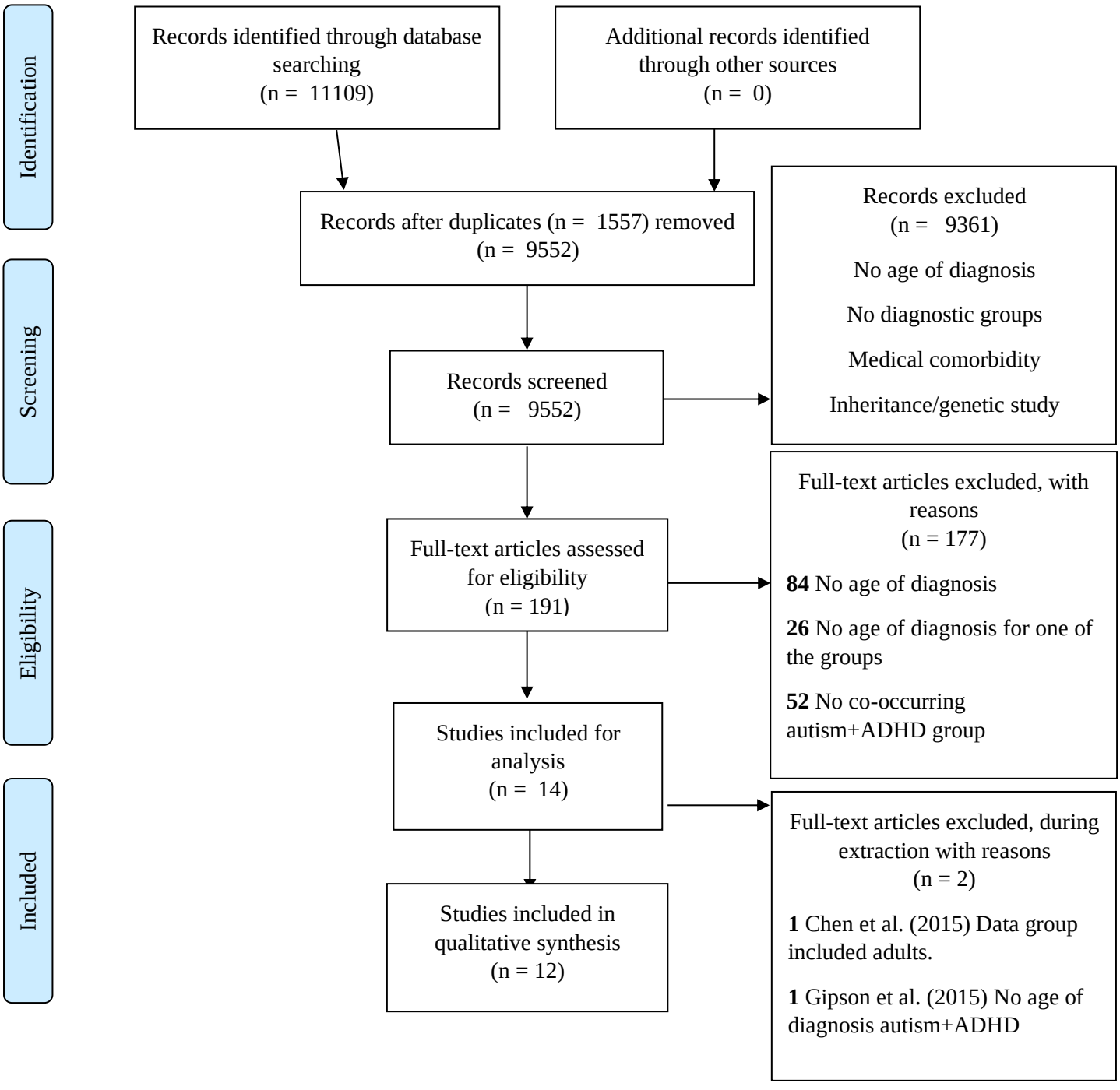
Community Participation

Community members were not involved in this study

Results

The PRIMSA flow diagram in *Figure 2.1* represents the study selection process (Moher et al., 2009). The initial screen yielded 11109 results. After the duplicates were removed, 9552 articles were screened and 9361 records were excluded. One-hundred-and-ninety-one full text articles were assessed for eligibility at the full text stage, and 177 were excluded due to: (i) not providing quantitative data on the age of diagnosis, (ii) not including a co-occurring autism + ADHD group, (iii) not having an age of diagnosis for at least two groups, (iv) no comparison with autism or ADHD only group or whole group, (v) a review or a brief report, or a case study or (vi) including an adult population. Two studies that were originally included in the extraction were then excluded due to closer examination revealing that an adult population had been included (Chen et al., 2015) and that age of diagnosis for the autism + ADHD had not been reported (Gipson et al., 2015).

Figure 2.1. PRISMA flow diagram



Participants

Table 2.1 summarises the demographic characteristics, location and diagnostic determination for the 12 included studies. These studies included a total of 41,382 children with autism, ADHD, or autism + ADHD diagnoses, of which 14,584 were diagnosed with autism only, 12,689 were diagnosed with ADHD only, and 14,109 were diagnosed with autism + ADHD. All 12 studies reported participant gender and the percentage of male participants ranged from 78.5% to 85.8%. Most studies were conducted in the USA, and studies were also conducted in Denmark, the Netherlands, Taiwan, Japan and China. The two studies each from Taiwan and The Netherlands used the same respective national databases (Kentrou et al., 2019; Lin et al., 2014; Wang et al., 2018; Wei et al., 2018). The participant data range was from 1995 until 2020 across all studies. The main method of diagnostic determination, or how the study ascertained the diagnoses of the respective children, was through medical records. The exceptions included Miodovnik et al. (2015), Stevens et al. (2016), and Wang et al. (2018) where their diagnostic determinations were done through surveys or interviews with parents. The participants were drawn from three categories described as population, clinical and database. Population included regional sampling of a population through general medical records across multiple health care settings, which was not specific to psychiatric hospitals or clinics specialising in diagnosis of either autism or ADHD. Clinical settings included any specific population drawn from a diagnosing clinic or specific psychiatric hospital. A database population was described as an elected database for parents to join although in some cases these databases were linked with access to nationally funded support.

Table 2.1.

Demographic characteristics of participants in the included studies.

Included studies	Total n. [a]	Total n. autism only	Total n. ADHD only	Total n. autism ADHD	Male %	Geographic location	Date range of data set	Diagnostic determination	Participant description
1. Engelhard et al., 2020	1658	343	1175	140	79	USA	2013-2020	Medical records	Population
2. Hatakenaka et al., 2016	65	42	14	9	84	Japan	2012-2013	Medical records	Clinic
3. Jenson & Steinhaus., 2015	8,958	NA	7116	1842	79.4	Denmark	1995-2010	Medical records	Clinic
4. Joshi et al., 2017	181	NA	74	107	82.5	USA	2005-2012	Medical records	Clinic
5. Kentrou et al., 2019	449	392	NA	57	80.2	Netherlands	Not given	Parent/self-report	Database
6. Lin et al., 2014	5130	498	4237	395	82	Taiwan	1995-2013	Medical records	Population
7. Miodovnik et al., 2015	1451	746	NA	705	82	USA	2011-2012	Parent report	Population
8. Soke et al., 2018	1091	785 (8yrs.)	NA	306	79.5	USA	2010	Medical records	Population
9. Stevens et al., 2016	1317	717	NA	600	78.8	USA	2011	Parent report	Population
10. Wallisch et al., 2018	363	259	73	31	80.7	USA	2000-2014	Medical records/survey	Clinic
11. Wang et al., 2018 (China)	433	186	NA	247	82.4	China	2013-2014	Parent report	Database
Wang et al., 2018 (Netherlands)	492	361	NA	131	78.5	Netherlands	2013-2014	Parent report	Database
12. Wei et al., 2018	19794	10255	NA	9539	84.8	Taiwan	2001-2011	Medical records	Population

[a] Reflects the total of the three groups, it does not include controls or other group totals in the studies.

Risk of Bias

The risk of bias of each study was assessed using the indicators outlined in Hoy et al. (2012) and the values are presented in Table 2.2. Nine studies were deemed low risk and four were deemed at moderate risk of study bias. The most common area of higher risk was in insuring that the participant pool was representative of a national sample. Studies that used parent report for both diagnosis and age of diagnosis had higher risk around case definition, because data were considered collected via proxy as opposed to medical records. All articles used the same mode of data collection, so were all low risk in this area

Table 2.2.

Risk of bias assessment results adapted from Hoy et al., (2012).

Risk of bias item [a]	1. Target pop.	2. Sample of target	3. Random selection	4. non-response bias minimal	5. data direct	6. Case definition	7. study instrument	8. mode of data collection	9. Prevalence period	10. No errors	Total high risk items.	Summary of overall risk [b]
1. Engelhard et al., 2020	No	Yes	Yes	Yes	Yes	Yes	N/A	Yes	Yes	Yes	1	Low
2. Hatakenaka et al., 2016	No	No	No	Yes	Yes	Yes	N/A	Yes	No	Yes	4	Moderate
3. Jenson & Steinhaus, 2015	Yes	Yes	Yes	Yes	Yes	Yes	N/A	Yes	No	Yes	1	Low
4. Joshi et al., 2017	No	No	Yes	No	No	No	N/A	Yes	Yes	Yes	5	Moderate
5. Kentrou et al., 2019	No	No	No	No	No	No	N/A	Yes	Yes	Yes	6	Moderate
6. Lin et al., 2014	Yes	Yes	Yes	Yes	Yes	Yes	N/A	Yes	Yes	Yes	0	Low
7. Miodovnik et al., 2015	Yes	Yes	Yes	Yes	No	No	N/A	Yes	Yes	Yes	2	Low
8. Soke et al., 2018	No	Yes	Yes	Yes	Yes	Yes	N/A	Yes	Yes	Yes	1	Low
9. Stevens et al., 2016	Yes	Yes	Yes	No	No	No	N/A	Yes	Yes	Yes	3	Low
10. Wallisch et al., 2018	No	No	Yes	No	Yes	Yes	N/A	Yes	No	Yes	3	Low
11. Wang et al., 2018 (China)	Yes	Yes	Yes	Yes	No	No	N/A	Yes	Yes	Yes	2	Low

Wang et al., 2018 (Netherlands)	No	No	No	Yes	No	No	N/A	Yes	Yes	Yes	5	Moderate
12. Wei et al., 2018	Yes	Yes	Yes	Yes	Yes	Yes	N/A	Yes	Yes	Yes	0	Low

[a] The following questions guided the assessment: 1. Was the study's target population a close representation of the national population in relation to relevant variables?; 2. Was the sampling frame a true or close representation of the target population?; 3. Was some form of random selection used to select the sample, OR was a census undertaken? 4. Was the likelihood of nonresponse bias minimal?; 5. Were data collected directly from the subjects (as opposed to a proxy)?; 6. Was an acceptable case definition used in the study? e.g. authors specify method by which ADHD or autism was diagnosed, such as ADOS using DSM V criteria; 7. Was the study instrument that measured the parameter of interest shown to have validity and reliability?; 9. Was the length of the shortest prevalence period for the parameter of interest appropriate?; 10. Were the numerator(s) and denominator(s) for the parameter of interest appropriate?

[b] Low risk 0-3; Moderate risk 4-6; High risk 7-9

Age of Diagnosis

Table 2.3 presents the differences in age of autism and ADHD diagnosis depending on whether individuals were diagnosed with autism, ADHD, or autism + ADHD. In Wang et al. (2018) the age of autism diagnosis for the autism + ADHD only group was compared to the age of autism diagnosis across the whole sample, rather than an autism only group. The age ranges for each study differed and varied from 0 year to 18 years across the studies. The studies of Hatakenaka et al. (2016) and Wallisch et al. (2020) were restricted to participants under the age of 6 years old. Soke et al. (2018) provided the mean difference between age of diagnosis for 8 year old children with autism + ADHD and autism only. Joshi et al. (2017) provided the age of onset between groups.

Age of Diagnosis for Autism

The average age of autism diagnosis ranged from 3.5 years (Hatakenaka et al., 2016) to 6.2 years (Lin et al., 2014). The average age of autism diagnosis when children had co-occurring ADHD ranged from 3.3 years (Wang et al., 2018) to 7.5 years (Lin et al., 2014). In the autism + ADHD group, the average age of autism diagnosis was between 0.7 (Wallisch et al., 2020) and 1.8 years (Kentrou et al., 2019; Miodovnik et al., 2015; Wei et al., 2018) later compared to the autism only group.

Age of diagnosis for ADHD

The average age of ADHD diagnosis ranged from 4.9 years (Wallisch et al., 2020) to 9.8 years (Jensen & Steinhausen, 2015). The average age of ADHD diagnosis when children had co-occurring autism ranged from 4.8 years (Hatakenaka et al., 2016) to 8 years (Jensen & Steinhausen, 2015). In the autism + ADHD group, the average age of ADHD diagnosis was between - 0.1 (Wallisch et al., 2020) to 1.8 years (Jensen & Steinhausen, 2015) earlier compared to the ADHD only group.

Table 2.3.

Mean age of autism and ADHD diagnosis and difference in age of diagnosis for autism only, ADHD only, and autism + ADHD groups

Included studies	Age range (years)	Mean age of autism diagnosis (years)			Mean [a] differ.	Sig.	Mean age of ADHD diagnosis (years)		Mean [a] differ.	Sig.
		Whole sample	autism only	autism + ADHD			ADHD only	autism + ADHD		
1. Engelhard et al., 2020	1 - 13		4.1(1.8)	5.5 (2.4)	-1.4		7.2(1.8)	5.9 (1.8)	+1.3	
2. Hatakenaka et al., 2016	1.9 - 6		3.5 (1.1)	4.8 (0.8)	-1.3		5.1 (1.1)	4.8 (0.8)	+0.3	
3. Jenson & Steinhaus, 2015	4 - 17						9.8 (3.5)[b]	8	+1.8	
4. Joshi et al., 2017[c]	6 - 17						4.0 (1.9)[c]	3.5 (1.7)[c]	+0.5	p = 0.12
5. Kentrou et al., 2019	4 - 18	5.7 (2.5)	5.3 (2.1)[b]	7.1 (2.4)[b]	-1.8	p <0.001				
6. Lin et al., 2014	0 - 18		6.2 (4.4)	7.5 (3.7)	-1.3	p < 0.001	8.4 (3.1)	7.4 (3.3)	+1.0	p < 0.001
7. Miodovnik et al., 2015	2 - 17		4.6 (0.2)	6.4 (0.3)[b]	-1.8	p < 0.001		5.9 (0.2)[b]		
8. Soke et al., 2018[d]	8				-1.0	p ≤ 0.05				
9. Steven et al., 2016[e]	6 - 17		4.7 (2.7)	6.3 (3.2)	-1.6	p < 0.001				
10. Wallisch et al., 2018[e]	3 - 6	4.5	4.3	5.0	-0.7	p < 0.001	4.9	5.0	-0.1	
11. Wang et al., 2018		5.0 (2.2)		5.2 (2.0)	-0.2					
(Netherlands)	6 - 14									
Wang et al., (China)	6 - 14	3.3 (1.1)		3.3 (1.2)	0.0					
12. Wei et al., 2018	0 - 17	5.9	5.1 (3.1)	6.9 (3.0)[b]	-1.8	p < 0.001		6.7 (2.8)[b]		

Note: Whole sample included all children diagnosed with autism including autism only and autism+ADHD.

[a] Mean difference for age of autism and ADHD diagnosis was calculated by subtracting the age of diagnosis for the autism + ADHD group from the age of diagnosis of the autism/ADHD only group, or the whole sample.

[b] Two data points combined using weighting for the sample because data in original study was split by gender or order of diagnosis (e.g. ADHD before autism/ADHD same/after autism).

[c] Mean age of onset for the diagnoses.

[d] Only the mean difference was provided

[e] Months converted to years.

Results of possible contributing factors to age of diagnosis

In the included studies some factors were quantifiably measured in comparison to the co-occurring group and the single diagnosis**.** Some studies included the influence of these factors in general on age of diagnosis, but did not report the impact separately on a co-occurring group so are not included in the results section. The factors that were quantifiably measured in comparison to a co-occurring group included age of parental first concerns; diagnostic wait times, order of co-occurring diagnosis, and presence of intellectual disability (ID), and the effect of gender on age of diagnosis.

Age of First Parental Concerns

Two studies compared the timing of when parents first had concerns for their children. Stevens et al. (2016) found that parents in the autism +ADHD group first expressed concerns that their child might have autism at 2.8 years compared to 2.4 years in the autism only group, a statistically significant difference ($p < 0.001$) . Stevens et al. (2016) also investigated whether there was a significant difference between parents' first concern and when medical assistance was sought, and found that help seeking was significantly later for the autism + ADHD group than the autism only group ($p < 0.001$). (Wang et al., 2018) found that parents in the autism + ADHD group first expressed concerns that their child might have autism at 2.4 years in China and 3.4 years in the Netherlands, compared to 2.5 years in China and 3.2 years in the Netherlands in the whole sample ($p < 0.05$). Joshi et al. (2017) was the only study which examined the age of parents first ADHD concern. They found that parents in the autism + ADHD group first expressed concern that their child might have ADHD at 3.5 years compared to 4.0 years for the ADHD only group, which was not statistically significant.

Diagnostic Wait Times

Two studies examined the difference in the time between a first visit to a practitioner to the

time at which the child is diagnosed. Hatakenaka et al. (2016) found that the autitsm + ADHD group waited 5 months longer than the autism only group for a diagnosis of autism after the first visit, and the ADHD only group waited 8 months longer than the autism group for a diagnosis (no statistical analysis performed). Stevens et al. (2016) found that the autism + ADHD group waited 1.3 years longer than the autism only group for a diagnosis after first seeking medical assistance, however statistical analysis was carried out for age of first concern to age of diagnosis and age of first concern to age first medical assistance sought but not medical assistance sought to age of diagnosis.

Order of co-occurring diagnosis

Miodovnik et al. (2015) and Wei et al. (2018) found that age of autism diagnosis was significantly later in children with autism + ADHD for whom ADHD was diagnosed first: 8.6 years (Miodovnik et al., 2015) and 8.5 years (Wei et al., 2018), in comparison to those who had their ADHD diagnosed at the same time or after their autism diagnosis: 4.7 years (Miodovnik et al., 2015) and 5.5 years (Wei et al., 2018) ($p < 0.001$ for both studies).

Presence of ID (intellectual disability)

Stevens et al. (2016) found that an additional diagnosis of intellectual disability (ID) led to a reduction in the age of diagnosis of autism when co-occurring ADHD was present from 6.3 years to 5.4 years. The autism diagnosis for the co-occurring with ID was still significantly later than an autism diagnosis of a child with autism + ID (3.7 years) or autism only (4.7 years) ($p < 0.001$). Hatakenaka et al. (2016), Jensen and Steinhausen (2015) Miodovnik et al. (2015), Soke et al. (2018), Wei et al. (2018), and Wallisch et al. (2020) discuss ID or developmental delays in relation to age of diagnosis or diagnosis under 6 years, but not in relation to the co-occurring group. Joshi et al. (2017) excludes children with an IQ under 70.

Gender

Kentrou et al. (2019) found that being female led to an increase in the age of autism diagnosis compared to being male. These gender differences in the age of autism were exacerbated with the co-occurring group being diagnosed 1.5 years later in males and 2.6 later in females compared to a gender differences in the autism only group (Kentrou et al., 2019) ($p < 0.001$).

Discussion

The primary aim of this systematic literature review was to examine the difference in age of autism and ADHD diagnosis for an autism + ADHD group in comparison to children with only one of these diagnoses. The 12 articles in this review supported the hypothesis that there is a significant delay in autism diagnosis with co-occurring ADHD. The delay in age of autism diagnosis ranged between 0.7 years and 1.8 years compared to an autism only group. The reverse trend was also apparent with children over six years, with ADHD being diagnosed earlier when co-occurring autism was present. The earlier age of ADHD diagnosis for children over 6 years ranged between 1 to 1.8 years compared with an ADHD only group. Quantifiable factors that may have impacted the timing of these diagnoses included slightly later parental first concerns for the autism + ADHD group compared to autism only and slightly earlier compared to an ADHD only group. Children with co-occurring autism + ADHD and their families also waited longer for a diagnosis. Age of diagnosis may also be affected if they received their ADHD diagnosis before the autism diagnosis or if the child is female. The impact of later parental concerns, gender, and order of diagnosis, must be interpreted with caution due to a scarcity of research looking at these factors in relation to the age of diagnosis across these diagnostic groups.

The inverse relationship, where an ADHD diagnosis is made 12 to 20 months earlier when autism is present over the age of six years, has not been a focus of the research

literature (Lin et al., 2014). The three studies that mentioned this earlier diagnosis of ADHD attributed this finding to greater symptom severity of autism + ADHD and earlier developmental identification associated with autism characteristics (Engelhard et al., 2020; Jensen & Steinhausen, 2015; Lin et al., 2014). Elwin et al. (2020) demonstrated that when autism co-occurred with ADHD, ADHD symptoms were quantifiably more severe than in an ADHD only population. Therefore, this severity might indicate that the ADHD-symptom profile is more apparent to caregivers and leads to earlier diagnosis. In the study by Joshi et al. (2017), however, the autism + ADHD group did not look different from an ADHD only group in terms of severity. In the two studies that included a population under six years the difference in age of ADHD diagnosis was not apparent (Hatakenaka et al., 2016; Wallisch et al., 2020). This might be due to the ADHD diagnosis being given simultaneously with autism and the inclusion of participants only under the age of six years when ADHD is usually diagnosed after the age of six (Visser et al., 2014). The effect of ID on the age of diagnosis of a co-occurring group also has mixed results. Stevens et al. (2016) was the only study to statistically report an earlier diagnosis effect on a co-occurring group with and without ID; however, there is some evidence that ID has the reverse or no statistically significant effect on the age of diagnosis across a combined autism group (Miodovnik et al., 2015; Wei et al., 2018)

Delays in diagnoses can occur at various points along the clinical pathway, from identification of early atypical development, to help-seeking behaviour, and a diagnostic assessment (Becerra-Culqui et al., 2018; Yamauchi et al., 2015). Two studies in the systematic review showed that the co-occurring group had slightly later parental concerns than the autism only group (Joshi et al., 2017; Stevens et al., 2016; Wang et al., 2018). Stevens et al. (2016) also showed that there was a delay between parents' first concerns and when they sought medical assistance. Although beyond the scope of this literature review,

one possible explanation is the nature of that parental concern. There was some evidence that the autism + ADHD group have parental concerns which are more likely to be behavioural in nature rather than the key signs of early autism diagnosis, such as language delay, social concerns, or fixed and rigid behaviour (Wallisch et al., 2020). Parents may not report a behavioural concern to their child's clinician until after the behaviours are seen as developmentally inappropriate, typically past the age of 6 years and when the child is in a school setting, which further delays help-seeking (Yamauchi et al., 2015; Zablotsky et al., 2017). The parental concerns of autism + ADHD children more closely resemble parental concerns of an ADHD group than an autism only group (Wallisch et al., 2020). This fits with Joshi et al. (2017) results which suggest that ADHD with co-occurring autism does not look significantly different in clinical manifestation from ADHD alone in measures such as type, or number of symptoms, diagnostic subtypes or even age of onset. An important future research direction would clarify patterns of parent referral concerns in relation to wait times to diagnosis.

After parental concerns and help-seeking comes the clinicians' role and subsequent diagnosis (Becerra-Culqui et al., 2018; Yamauchi et al., 2015). Greater wait times between first seeking medical assistance and wait time to diagnosis was reported between 0.4 to 1.3 years later than an autism only group than an autism + ADHD group (Hatakenaka et al., 2016; Stevens et al., 2016). There is evidence that children with autism + ADHD see a higher number of professionals prior to diagnosis and spend longer on wait lists prior to getting a diagnosis (Eggleston et al., 2019). ADHD symptoms may also mask autism symptoms during clinic assessments. (Gipson et al., 2015; Soke et al., 2018; Stevens et al., 2016). Nomura et al. (2014) studied children with autism at age 5 years who had shown signs of ADHD and found that hyperactivity and inattentiveness were apparent during a short visit, but subsequent parental home reports and observations during school revealed more autism symptoms

(Nomura et al., 2014). It has also been suggested that "search satisfying" occurs, where by a clinician finds one diagnosis that fits, and this leads to the delay in the recognition of autism symptoms and diagnosis (Gipson et al., 2015; Soke et al., 2018). This fits with the findings that the delay in autism diagnosis increases when the population of autism + ADHD children is divided into children that received their ADHD diagnosis first and those that received their autism diagnosis first or at the same time as their ADHD diagnosis (Kentrou et al., 2019; Miodovnik et al., 2015; Wei et al., 2018). Kentrou et al. (2019) only included children in the autism + ADHD group if they had been diagnosed with ADHD first. Therefore, it is suggested that once a diagnosis is given, clinicians and parents may not seek further diagnostic clarification. An important future research direction would examine the role of the clinician in possible delays, including the different professional groups of clinicians, which might use different diagnostic methodology. Future research might also consider the impact specialised and non-specialised professionals in diagnostic delays and possible amelioration of this delay with specific training in diagnosing autism and co-occurring conditions.

There is also a well-known delay in identifying females on the spectrum, perhaps due to better camouflaging of social skills (Lai et al., 2017; Wood-Downie et al., 2020), but this delay is compounded by the addition of ADHD as it could be argued that the presentation further complexifies. Evidence for this complexity was given by Kentrou et al. (2019), who showed that the delayed autism diagnosis with co-occurring ADHD stretched out a further year when the child was female in comparison with male children with autism + ADHD. The more complex a diagnostic picture, and the less typical symptom profile, the more difficult the diagnosis (Avlund et al., 2020; Smith et al., 2019).

There were methodological issues with the studies included within this review. The assessment of bias (Hoy et al., 2012) demonstrated the most common areas of risk were a nationally representative sample, direct data collection and case definition. High-risk in the

latter two categories reflected a different methodology whereby parents were interviewed or surveyed. The age of diagnosis was recalled, rather than contemporaneously noted in medical records and parents defined the case definition by affirming that their child had been given a diagnosis. However, there is a potential limitation with medical records too, whereby different procedures for diagnosing autism and ADHD within and between countries and different clinicians responsible for diagnosing might impact the age of diagnosis. The fallibility of memory might limit the comparison across studies that used medical records (Kentrou et al., 2019; Miodovnik et al., 2015; Stevens et al., 2016; Wang et al., 2018). Miodovnik et al. (2015) addresses this concern citing both ADHD research showing parent-reported survey data was similar in estimates to insurance data (Visser et al., 2013) and equally consistency in parent reported autism diagnosis with two nationally representative surveys (Control & Prevention, 2006), concluding that there is convergent validity between parent reported diagnosis of autism and ADHD and other forms of data collection. However, this remains a limitation even if the consistency across studies suggests that whether recalled or collected contemporaneously in medical records there is a delay in autism diagnosis when ADHD is present.

In addition, the two studies from the Netherlands and the two from Taiwan each drew from the same nationally representative database; the Netherlands Autism Registrar and Taiwan National Health Insurance database, respectively. It is possible that the same participant pools were used across these studies, thus reducing the true participation numbers. A further limitation was the lack of reporting around subtypes (inattentive, hyperactive or combined) of ADHD present with autism. Of the 12 studies, only Joshi et al. (2017) differentiated subtypes of ADHD co-occurring with autism and this might be an additional factor to consider in future research on the age of diagnosis. In addition some studies controlled for differences in age of diagnosis, such as adjusting for child sex, race/ethnicity,

maternal education and study site, which reduced the delayed age of diagnosis for the autism + ADHD group by one month compared to unadjusted diagnosis delay of 1 year (Soke et al., 2018). Other studies did not control for these possible differences, which might have impacted on the age of diagnosis.

The current review was limited as the inclusion criteria specified articles would be published after 2014 to minimise difficulties posed by the DSM restrictions on dual diagnosis of autism + ADHD prior to 2013. Some articles published after 2014 included a data set range which was collected prior to 2013. However, diagnosis of co-occurring ADHD with autism was prevalent before 2014 because of the ethical responsibility of clinicians to provide treatment for ADHD. Due to research and specifically effective interventions for ADHD, clinicians have had the advice that they should make the co-occurring diagnosis when ADHD symptoms reach clinical significance (Goldstein & Schwebach, 2004). It is difficult to speculate on how much impact the DSM IV had on the restriction of a co-occurring diagnosis in these earlier populations. It is likely that increased acceptance of a co-occurring diagnosis will result in increased diagnosis of more moderate ADHD characteristics with autism, but the same trends in diagnosis were seen pre- and post-2014.

Another consideration is that studies can report age of diagnosis as an incidental finding, and although the literature review search terms were broad, it is possible that an article was missed. Furthermore, we used Hoy et al. (2012) as a risk of bias, and another indicator tool might have produced different results.

Implications

The findings of this review suggest that professionals should be aware of delay in autism diagnosis with co-occurring ADHD. A short visit with clear ADHD symptoms and parental concerns about behaviour should not rule out the presence of autism, and an ADHD diagnosis should not preclude further autism investigation. This review also highlights that children

with autism + ADHD with relatively equal “severity” of symptoms do not appear to present early to clinic compared to children with autism only. This diagnostic picture can be further complicated by the child being female or having been diagnosed with ADHD first, which might create diagnostic overshadowing. A delayed autism diagnosis is not unique to a geographical location. Therefore, being aware of this phenomenon will help reduce delayed and missed diagnosis. The studies examining contributing factors to this delay are few, but greater evidence of these factors would mean implications for screening, and assessment. Children showing factors associated with a delayed diagnosis might be subject to earlier screening and more in depth assessment.

There are several important directions for autism + ADHD research. Further examination of factors which may impact the age of diagnosis is warranted, such as which autism symptoms might be more apparent in short clinic visits. A greater detailed study of the pathway from parental concerns to help-seeking and diagnosis would help to identify which, if any, of these steps contributes most to delayed autism diagnosis when ADHD is present. Further research on the impact of factors such as later parental concerns, gender, order of diagnosis, and intellectual disability is also required to corroborate the results of the few studies which investigated these factors. More research into the effect of autism on early ADHD diagnosis would also be illuminating, particularly as severity and earlier developmental autism symptoms do not appear to be adequate explanations of an earlier diagnosis of ADHD. Another area for consideration in understanding delayed diagnoses, is an examination of delayed or missed adult diagnoses.

The findings of this review, that autism is typically diagnosed later when ADHD symptoms are present, and ADHD is typically diagnosed earlier when autism is present, are of concern. This is because an early diagnosis of autism is essential in enabling the provision of early intervention. A more complex diagnostic picture with less obvious key symptoms for

a diagnosis of autism might lead to longer waiting periods between the first clinic visit and an eventual diagnosis. Further research is needed to understand the weight and importance of each factor in the delay of an autism diagnosis and the earlier diagnosis of ADHD when the two conditions co-occur.

CHAPTER 3

OVERVIEW OF THE PRESENT RESEARCH

Project overview

Rationale and Aims for the Proposed Research

Timely and accurate diagnosis enables autistic and ADHD children to access early support. This can lead to more positive outcomes for those children (Whitehouse et al., 2021). The early years of life are when the brain is most plastic and attuned to learning language and social skills, which means that support may have more efficacy in improving cognition, language and social skills, as well as improving independence in the long-term (Franz & Dawson, 2019). However, there is a delay in diagnosing autistic children who also have significant ADHD characteristics, which has implications for accessing support during the early developmental period (Franz & Dawson, 2019) . This critical age for effective support requires that the relationship between ADHD co-occurrence and late diagnosis be better understood. Furthermore, the outcomes for children with both diagnoses are shown to be worse than either diagnosis alone, suggesting that this is a particularly high-need population for effective early support (McDougal et al., 2020; Rao & Landa, 2014; Yerys et al., 2019). Understanding why there is a significant delay in diagnosis is of great clinical importance, and in need of further research.

There is a growing body of research suggesting there is a delay in the diagnosis of autism in autistic + ADHD children. (Davidovitch et al., 2015, Eggleston et al., 2019; Kentrou et al., 2019; Miodovnik et al., 2015; Stevens, Peng & Barnard-Brack, 2016), however to date, there is relatively little research evaluating the reasons behind this delay. A number of theories have been put forward as to why there is a significant delay

in an autism diagnosis when ADHD is present (Miodovnik et al., 2015; Soke et al., 2018). One possibility is the timing and nature of a parent's first concern. When and what parents first view as a developmental concern can lead to either early, or late, seeking of professional advice. Stevens et al. (2016) conducted the only study to compare the timing of parental concerns between autism, and autism + ADHD groups. They conclude that both the first reported parental concern and the age at which a developmental assessment was sought were later for an autism + ADHD group of children than for those in an autism only group (Stevens, Peng and Barnard-Brak, 2016).

However, there is no research examining the nature of parental first concern and if it relates to the characteristics associated with the criteria for the subsequent diagnosis. There are also questions about whether these first parental concerns are different for parents of autism +ADHD, autism only, or ADHD children. Parental first concern is conceptualised as the concern which parents first presented to a professional. This is distinguished from the specific types of parental-observed atypical development which might have been apparent to parents, prior to them seeking help. Patterns of atypical development is also an under studied area for these groups. The study of atypical development and presenting parental concern could help to identify patterns of development that might differ across the diagnostic groups.

Therefore, it is not clear whether there is a pattern to the nature of the first parental concern expressed about a child with autism +ADHD characteristics, and whether this might reflect one condition or a combination of both conditions. Therefore, it is possible that the delay in diagnosis could be due, in part, to parents of autistic + ADHD children having later and/or different parental concerns than parents whose

children receive either diagnosis alone. If parents of autism + ADHD children have categorically different concerns, it is possible that they would not flag an activation of either diagnostic process. Therefore, in combining both hypothetical possibilities, parents of autistic + ADHD children might access clinical support later with a less clear referral concern than an autism or ADHD only population.

Another possible mechanism for the delayed diagnosis of autism + ADHD children is the role of clinicians and the diagnostic process (Davidovitch et al., 2015, Miodovnik et al., 2015). There are several possibilities as to why clinicians may be delayed in diagnosing autism. They are as follows: i) clinicians may be more familiar with diagnosing ADHD; ii) ADHD symptoms may be more salient during a clinical visit; iii ADHD symptoms may mask autism symptoms; iv) Clinicians may misdiagnose autistic children with ADHD; or v) autism + ADHD may have a mismatched developmental trajectory with symptoms, such as inattention and hyperactivity, more prominent at a younger age, than autism symptoms (Davidovitch et al., 2015, Kentrou et al., 2019; Miodovnik et al., 2015; Soke et al. 2018). Therefore, understanding how a clinician, who might be either a psychologist, psychiatrist, or paediatrician, conceives of the diagnostic complexity of an autism + ADHD child and issues around co-occurrence will shed further light on the possible reasons for the delay.

The diagnostic complexity of neurodevelopmental conditions, such as autism and ADHD are due to the increase likelihood of co-occurrence compared to a neurotypical population (Jensen & Steinhausen, 2015; Soke et al., 2018). The number of other conditions, which statistically are more likely to co-occur with autism and ADHD than a non-clinical population, are not only other neurodevelopment diagnoses, such as dyslexia and intellectual impairment, but also other physical, genetic and related

conditions, such as sleep, sensory processing issues, gastrointestinal difficulties and epilepsy (Jensen & Steinhausen, 2015; Neumeyer et al., 2019; Soke et al., 2018). All of these additional issues have the potential to mask, overshadow, or substitute for the possibility of correct diagnosis of autism or ADHD (Heyman et al., 2022). In addition there are some common characteristics shared across diagnostic categories and also heterogeneity in symptom profile expected within diagnostic categories (Uddin et al., 2019). This means that there are many behavioural characteristics that are not uniquely indicative of one specific diagnosis. When diagnostic case complexity occurs, there are often social difficulties and atypical attention characteristics as part of the complex profile (Heyman et al., 2022). For a child to fulfil the criteria of autism + ADHD, the child must be presenting with multiple complex traits, which are not better explained by a single diagnosis. Therefore, this complexity, plus the statistical likelihood of an additional condition or trait, means that to study autism + ADHD is to study case complexity.

A greater understanding of this diagnostic pathway might help explain why autistic + ADHD children have a significant delay in an autism diagnosis and access to intervention. This pathway includes parents noticing atypical development in their child, the timing and nature of parents' first presenting concern to a clinician, and their overall experience of the diagnostic pathway. It also includes understanding the diagnostic process from the point of view of clinicians, when they are faced with case complexity. Further, while there is limited investigation of the presence and cause of the delay in diagnosis internationally (Davidovitch et al., 2015; Kentrou et al., 2019; Miodovnik et al., 2015; Soke et al., 2018), there is extremely little in New Zealand.

Diagnosis of Neurodevelopmental Conditions in New Zealand

In New Zealand diagnosis of neurodevelopmental conditions can take place through a public or a private route (Drysdale & van der Meer, 2020). The public route usually involves a referral from general practitioner (GP) or, less commonly, from another professional within the field for a developmental assessment. There are two avenues for this assessment, which are loosely age related. Pre-school children are usually referred to a child development multidisciplinary team at an outpatient clinic within Te Whatu Ora (Health New Zealand). This was formerly the local District Health Board (DHB). There were 20 District Health Boards around New Zealand, which were seen as duplicating systems and promoting regional over national equality, so Te Whatu Ora (Health New Zealand) centralised this system in July 2022. The team at the hospital follows the guidelines outlined by the Ministry of Health and Education in New Zealand around diagnosis (Whaikaha, 2022). These guidelines stipulate that the diagnostic team should include two to three professionals, across multiple disciplines, but usually including either a paediatrician or psychiatrist, who assess using observations, questionnaires, developmental interviews and validated and reliable diagnostic tools (Drysdale & van der Meer, 2020). At primary school level this clinical setting is also used but often in conjunction with the child and adolescent mental health services (CAMHS), which follow similar guidelines with multidisciplinary teams (Thabrew & Eggleston, 2017). In adolescence, CAMHS appears to diagnose more adolescents than the Te Whatu Ora teams (Thabrew & Eggleston, 2017). After diagnosis, the child and family are usually referred to a Developmental Services Co-ordinator, which is a position to help support families post diagnosis (Drysdale & van der Meer, 2020).

The long waitlists for the public diagnosis provided by Te Whatu Ora and CAMHS have prompted a number of private clinicians, who can diagnose within their scope of practice, to offer an alternative system (Drysdale & van der Meer, 2020). Private clinicians are able to refer back to the public system at any point, so a hybrid private-public service is also possible (Drysdale & van der Meer, 2020). However, it is estimated that as few as 5% of autistic children in New Zealand are diagnosed privately (Thabrew & Eggleston, 2017). No estimates have been given for the percentage of ADHD children diagnosed privately. In both the public and private sector, diagnosing clinicians have identified lengthy wait times, lack of access to professionals in multidisciplinary teams, and lack of post diagnostic supports as particular areas of system failure (Thabrew & Eggleston, 2017).

A study of New Zealand parents' experiences of autism diagnosis for their child showed that parents had the highest satisfaction with the process when they had shorter wait times, had a multidisciplinary assessment, and received a written report (Eggleston et al., 2019). Lower satisfaction was associated with having a co-occurring diagnosis first (Eggleston et al., 2019). Families with an autistic + ADHD child spent longer on a wait list and saw more professionals, but there was no data gathered about a delay in diagnosis compared to autistic children in New Zealand. There is also poor understanding both in New Zealand and internationally about the possible causes of this delay. There is also little understanding internationally about whether the inverse occurs with an earlier ADHD diagnosis when autism is also present (Jensen & Steinhausen, 2015).

Research Questions

The research aimed to understand the patterns and possible interactions between the diagnosis of autism and ADHD, and by extension, complex cases. This was approached from a biopsychosocial position involving multiple perspectives of parents and clinicians. Both quantitative and qualitative analysis were utilised: Quantitative analysis to map the delay of autism diagnosis with co-occurring ADHD and explain possible contributing factors; and qualitative analysis to understand the phenomena of diagnosis of these conditions. Based on this rationale, the proposed research aimed to answer the following key questions:

Study 1

The following questions were examined via an anonymous survey of New Zealand parents who had children diagnosed with autism only, ADHD only, and autism + ADHD (See Chapter 4). This study aimed to quantitatively understand why there might be differences in the timing of both autism and ADHD diagnosis when the two conditions co-occur, compared to when they are present in isolation.

Q1. Is there evidence for timing differences in diagnosis when autism + ADHD co-occurs compared to a populations with only one of those diagnoses?

Q2. What factors might influence the age of ADHD and autism diagnosis?

- Does parent reported atypical development at particular ages influence age of diagnosis?
- Do certain presenting concerns to a health professional trigger a quicker diagnosis pathway?
- Does the co-occurring autism + ADHD group seek specialist help later and see more specialist prior to diagnosis than other diagnostic groups?

Q3. Are children with autism + ADHD similar or different in their developmental trajectory and help-seeking behaviour than children with ADHD only or autism only?

Study 2

The following question was answered by a qualitative section of the anonymous survey of New Zealand parents who have children diagnosed with autism only, ADHD only and autism + ADHD (See Chapter 5). This study aimed to qualitatively understand parents' experience of obtaining a diagnosis for their child. This might provide further insight into possible factors contributing to a delayed autism diagnosis and thematic patterns across and between the diagnostic experience.

Q4. What can parents' perception of the diagnostic process for either autism, ADHD or autism + ADHD add to our understanding of the comparative experience and possible delays?

Study 3

The following questions were answered by a sample of New Zealand clinicians (paediatricians, psychologists and psychiatrists) responding to two vignettes describing complex cases in an anonymous survey (See Chapter 6). The first vignette described a hypothetical 4-year-old and the second described a hypothetical 8-year-old, who both presented with neurodevelopmental case-complexity, including a number of autism and ADHD features. This study aimed to understand how clinicians approached complex cases and if there was a pattern to diagnosis. This clinicians' perspectives might help identify further aspects where delays in diagnosis might occur.

Q5. What assessments, hypothetical diagnoses, and support services do clinicians recommend for complex co-occurring neurodevelopmental cases?

Q6. Is there a consistent pattern or pathway recommended for complex co-occurring neurodevelopmental cases?

Research Hypotheses

In line with the above questions and literature, the hypotheses were as follows:

Study 1.

1. Parents of autistic + ADHD children will describe later concerns, combined with a later diagnosis, compared to parents of children with autism-only. However, existing research does not give an indication as to whether or not there is a pattern to the type of parental first concern.
2. The autism + ADHD children will more closely resemble a pattern seen in the ADHD children's development and help-seeking, due to these children often receiving a late diagnosis of autism and an ADHD diagnosis sooner.

Study 2.

3. There will be common themes and themes unique to each diagnostic groups, and parents will express particular points in the diagnosis process that were pertinent to their experience.

Study 3.

4. Clinicians will have a common preferred pathway for the assessment of these complex-cases.

5. The inclusion of ADHD characteristics in the vignettes will lead to the clinicians' favouring an ADHD diagnosis.

Theoretical Framework

The author of this thesis takes the position of biopsychosocial model of autism + ADHD acknowledging biological, psychological, social factors and macro-cultural

variables are needed to conceptualise an understanding of delayed diagnosis and access to support (Suls & Rothman, 2004). A transdisciplinary approach aims to explore the issue from parents, system for diagnosis, and clinicians' perspective without assigning 'fault' for delay in diagnosis (Suls & Rothman, 2004).

Research Design

Studies 1 and 2 use a non-experimental questionnaire design aimed at gathering information on diagnostic issues pertaining to children who have been diagnosed with autism only, ADHD only and autism + ADHD. Study 3 used a professional-judgement report questionnaire based on hypothetical vignettes of complex cases designed for clinicians involved in the diagnosis of neurodevelopmental conditions. Both questionnaires were designed to be anonymous online surveys, and included a mix of quantitative and qualitative questions. These surveys aimed to capture multiple perspectives by targeting a range of participants. Biological components were examined in questions related to atypical development and clinical assessment processes and environmental factors such as timing and the quantities of types of specialists. There were questions related to psychological and social components of parents' and clinicians' experience and thus biopsychosocial model was used to answer questions on the diagnosis of autism, ADHD and complex cases.

A mixed method design is where both methods are used to simultaneously garner a more complete understanding of diagnosis, described as "convergent mixed-method" (Creswell & Hirose, 2019). Meyer and Schutz (2020) discuss that mixed-method design increasingly being used in the discipline of educational psychology due to the interdisciplinary nature of the field and the ability of mixed methods to capture multiple worldviews. Multiple views, both quantitatively and qualitatively examined,

can help reconcile inequities in education and health by increasing the voices that are heard (Meyer & Schutz, 2020). Quantitative components provided an understanding of the timing and factors impacting diagnosis, while the qualitative components enabled an examination of the phenomenon of the experience of getting a diagnosis (Creswell & Hirose, 2019). The research design of this thesis included quantitative and qualitative data analysed and discussed separately within each study before being examined together in the final chapter of this thesis.

Data Collection

The surveys were anonymous which has shown to be advantageous when asking sensitive questions, in that parents and clinicians might feel more confident to answer with frank honesty (Ong & Weiss, 2000). Some of the initial concerns around online surveys not representing a wider demographic of parents (Heiervang & Goodman, 2011) has been reduced, particularly with national Covid-19 lockdowns promoting equity to online access (Mahoney et al., 2021). However, concerns around who has time and access to online surveys and issues of demographic reach remain a limitation of using surveys .

The questionnaire for Study 1 and 2 (see Appendix B) began with demographic questions, followed by quantitative questions pertaining to atypical development, first concern and age of diagnosis. The independent variables were diagnostic group (autism, ADHD, or autism + ADHD). The dependent variables were: i) the number and type of parent-reported areas of atypical development in the first year of life; ii) the age at which parents felt specialist consultation was needed; iii) the behaviour that led to specialist consultation and whether this aligned with the child's subsequent diagnosis; iv) the wait time from when parents felt specialist consultation was needed to when a

diagnosis was given; v) the types of specialist seen before a diagnosis; vi any additional diagnoses; and vii) parental agreement that their child's behaviour matched their diagnosis. The questions were based on a survey designed for the Australian Autism Biobank Family Questionnaire (Alvares et al., 2018) and the atypical development options were based on previous research (Waddington et al., 2022) (see Chapter 4).

The qualitative questions in Study 2 were designed to be open-ended with a wide angle lens that avoided judgment by asking what was helpful/unhelpful and finally, asking what participants would like researchers to know (Braun et al., 2020). The latter question was a way to demonstrate genuine curiosity and undermine a 'sense of agenda' often criticised in qualitative online surveys (Braun et al., 2020). This question also created a power shift for the participant to see themselves as having valuable knowledge or lived experience, which can motivate participants to explain their perspective (Harris et al., 2020).

Some researchers question the use of qualitative anlaysis of data collected from surveys (Braun et al., 2020; Creswell & Hirose, 2019). Criticisms include that the data produced is not rich enough to warrant qualitative analysis, it does not allow for follow-up or probing, and that a qualitative survey, if used, must be embedded solely in qualitative values without mixing any quantitative data collection (Braun et al., 2020). However, the survey was structured in such a way as to encourage engagement in the open ended qualitative responses. This included the survey starting with short answers first which explored timing, first concerns etc., so that parents were prompted by these answers to remember the diagnostic process and parents were shown a 90% completion rate on the completion-percentage-tab before the last three open-ended questions (Saleh,

2017). The open ended questions were also formulated with maximum clarity and after parents had shared a number of quantitative details (Lenzner et al., 2010) .

Data Analysis

Quantitative statistical analysis was used to understand if there were significant differences between the three groups across the measured variables. Excel was used to order the data exported from the Qualtrics surveys. The ordering included excluding participants who had not reached criteria and turning dichotomous data into either presence (1) or absence (0) thereof. Some coding required inter-rater agreement, such as coding for the nature of the presenting concern fitting with diagnostic criteria for a particular diagnosis.

The range of data necessitated the use of a range of statistical tests. The age of diagnosis was not normally distributed and required a Mann-Whitney-U test to compare across two groups. Kruskall-Wallis tests were used to compare differences for all other continuous variables. Chi-squared tests were used to compare differences in the demographic data and in expected relationships, such as the number of additional diagnoses and number of types of specialist seen, across the three diagnostic groups. Due to multiple comparisons, a Bonferonni correction was used for post-hoc tests by multiplying all p values by 3.

Data analysis of the three qualitative questions with open text responses resulted in a qualitative thematic analysis approach. Braun and Clarke (2006) have provided a guide in psychology for the thematic analysis approach, however, more recently they have refined this technique with the term "reflexive thematic analysis". This new form of thematic analysis is different from how Braun and Clarke (2006) originally articulated thematic analysis in psychology. Reflexive thematic analysis has an added

emphasis on "reflexivity" of the researcher. This entails a more lengthy period of "sitting with the data" and moving away from interrater reliability of coding to acknowledge that subjectivity and a non-linear progression of analysis is inherent in reflexive thematic analysis. The answers to the three questions, i) “What was helpful?,” ii) “What was unhelpful?,” iii) and “What else would you like a researcher to know?,” were treated as discreet individual text responses. This included attaching identifiers of diagnosis to each response, such as (autism) or (autism + ADHD). Descriptive statistics (percentages) were used to show how many participants did not respond and of those who did, approximately how many words they wrote.

The Study 3 questionnaire (See Appendix C and Appendix D) began with demographic questions followed by a vignette of a complex case for a four and eight year-old. The same questions were asked after each vignette. The small number of clinicians meant that descriptive statistics were used to analyse the results. Descriptive statistics were used to illustrate the sample demographic characteristics, the median level of concern across clinicians for each referral behaviour, and the percentage of clinicians who suggested different possible assessments, diagnoses, and support services. Specific named assessments were coded into categories. Descriptive statistics were also used to examine assessments, diagnoses and services that were commonly suggested together. The concerns listed in the vignette referral were also matched to diagnostic criteria using inter-rater reliability.

Ethical Considerations

Ethics approval for Studies 1, 2 and 3 was obtained by the Victoria University of Wellington Human Ethics Committee [Approval number 28993]. Informed consent was achieved through the first page of the anonymous survey specifying the purpose, the

aim of the project, the procedure for withdrawal of participation, and what will happen to the information participants give. The first page also provided contact details in the event of questions or problems and the specifics of the Human Ethics Committee information and approval number. Anonymity in all surveys was ensured by no direct identifying information being requested and any accidental material, such as names of schools or organisations, being removed.

An ethical challenge of Study 3 was that the initial explanation about the research could not include autism and ADHD as it could bias the answers of clinicians. Therefore, there was deception involved with the initial explanation of the purpose of the study. The introduction to the study explained my research interest as diagnosing complex behaviour presentations of school-aged children. At the conclusion of the survey, participants will be debriefed about my research investigating autism and ADHD co-occurrence. It was explained that although my research was in these areas, the vignettes did not represent an exact example of this diagnostic profile and could represent diagnostic complexity more generally.

Another ethical consideration relates to inequities and systemic racism in education and health. While it is heartening that Māori appear to have been well-represented in the research, this research did not follow a kaupapa Māori research-policy or directly address diagnosis of a neurodevelopmental conditions for Māori (Hikaka et al., 2022). An online survey also has the challenge of targeting a higher socio-economic population, who have digital access inequities. The survey's demographic in terms of income and education was representational of New Zealand population spread according to OECD data on New Zealand (OECD, 2019, 2022) (see chapter 5).

Other ethical considerations can occur in the reflexive thematic analysis. In analysing text, voices of participants, who wrote more and had more time to participate might be prioritised over others. Reflexive thematic analysis hopes to ameliorate some of these ethical concerns by encouraging multiple voices to be heard and the reflexivity of the researcher to be analysed by repeated examinations of the data and time to "dwell with" the data (Braun & Clarke, 2021).

CHAPTER 4

STUDY 1: Parent-reported early atypical development and age of diagnosis for children with co-occurring autism and ADHD

Chapter Note

This chapter was published as a journal article in the *Journal of Autism Developmental Disorders* titled *Parent-reported early atypical development and age of diagnosis for children with co-occurring autism and ADHD*. Dr Hannah Waddington helped with the design, analysis and editing. Prof. Andrew Whitehouse and Dr Kelly Carrasco assisted with editing and design. I designed, conducted and wrote this article and it is entirely my work aside from the input specified above. Aside from changes to ensure consistency of formatting with the rest of the thesis, this chapter is identical to the published article. The complete citation for the published article is: Sainsbury, W.J., Carrasco, K., Whitehouse, A.J.O., Waddington, H. (2022) Parent-reported Early Atypical Development and Age of Diagnosis for Children with Co-occurring Autism and ADHD. *Journal of Autism and Developmental Disorders,* 1-12. https://doi.org/10.1007/s10803-022-05488-0

Abstract

Autism and attention-deficit/hyperactivity disorder (ADHD) often co-occur. This survey of 288 New Zealand parents of children diagnosed with autism (n = 111), ADHD (n = 93), or both conditions (n = 84), examined the relations between age of diagnosis and early atypical development, the age specialist consultation was needed and types of specialists seen. Co-occurring autism and ADHD was associated with an earlier ADHD diagnosis and a later autism diagnosis. Parents of children with both diagnoses reported

less atypical development in language and social behaviours compared to parents of children of autism, and this co-occurring group also experienced longer wait times to diagnosis, and saw more types of specialists prior to a diagnosis, than those with autism.

Keywords

Co-occurring conditions, diagnosis, autism, attention deficit hyperactivity disorder, age of diagnosis, atypical development.

Autism spectrum disorder (hereafter referred to as autism) and attention-deficit/hyperactivity disorder (ADHD) are both neurodevelopmental conditions. Autism is characterised by impairments in social communication and interaction and rigidity of behaviour (American Psychiatric Association, 2013). Autism can be diagnosed at 1.5 years of age, but a meta-analysis found autism is usually diagnosed around 5 years of age (van't Hof et al., 2021). ADHD is defined by a triad of core symptoms that include inattention, impulsivity, and hyperactivity which impairs a child's functioning across multiple environments (American Psychiatric Association, 2013). In the United States of America (USA), ADHD is usually diagnosed around 6 years (Visser et al., 2014). It is estimated that anywhere between 14-78% of autistic children also have ADHD (Mannion & Leader, 2013), making this one of the most common co-occurring psychiatric conditions (Antshel et al., 2016). The wide ranging estimates of co-occurrence may be attributed to differences in study population samples and whether researchers were measuring symptoms of ADHD or official clinical diagnoses (Mannion & Leader, 2013).

The study of co-occurring conditions is in its nascence. Multiple new models or approaches have been suggested, to conceptualise co-occurrence beyond discrete

disease categories (Forbes et al., 2016; Rosen et al., 2018). In addition to a theoretical position, the study of autism + ADHD was not given attention because the Diagnostic and Statistical Manual of Mental Disorders (DSM) excluded the dual diagnosis of these conditions until the 5th edition (Kentrou et al., 2019).

Children with autism + ADHD often benefit from early intervention, which can decrease behavioural challenges and barriers associated with the conditions, and increase wellbeing and quality of life (Franz & Dawson, 2019; Risley et al., 2020). Access to early intervention during a sensitive developmental period can be hindered by a delay in diagnosis. Indeed, the presence of co-occurring ADHD may lead to a later diagnosis of autism compared to autism in the absence of ADHD (Jónsdóttir et al., 2011; Soke et al., 2018). Miodovnik et al. (2015) found that children with co-occurring autism and ADHD who received their ADHD diagnosis first, waited an average of 3 years longer for the additional diagnosis of autism. This was consistent across childhood and independent of the severity of symptoms. Wei et al. (2018), in a study involving 9,539 children with autism + ADHD, demonstrated that, regardless of the order of the diagnoses, (i.e. ADHD first or autism first), the diagnosis of autism was delayed by an average of 1.8 years compared to the age of diagnosis in an autism only population.

There is comparatively little research into the age of ADHD diagnosis for children with autism + ADHD compared to those with ADHD only. Research suggests children with both diagnoses receive their ADHD diagnosis between 1-1.8 years earlier than ADHD only children (Sainsbury, Carrasco, Whitehouse, McNeil, et al., 2022). This may be because autism is developmentally apparent earlier and thus co-occurring ADHD could be diagnosed sooner due to earlier help-seeking (Jensen & Steinhausen, 2015).

A greater understanding of the delay in autism diagnosis and early diagnosis of ADHD with co-occurring autism + ADHD requires an understanding of the diagnostic pathway for families. Previous research has found that parent-reported concerns about their child's development between 1-2 years of age were predictive of an early autism diagnosis (Guinchat et al., 2012; Ozonoff et al., 2014). In particular, observed atypical development in language and social communication predicted earlier diagnosis (Ozonoff et al., 2014). In ADHD research parent-reported symptoms at 3 years predicted the diagnosis of ADHD at the expected age of 6+ years (Oerbeck et al., 2020). However, little is known about the effect of parent-reported atypical development in the first year of life on age of diagnosis for children diagnosed with autism + ADHD compared to autism or ADHD alone.

After noticing atypical development, parents often present their concerns to a specialist (Zablotsky et al., 2017). Waddington et al. (2022) found that, for children with a diagnosis of autism, the age at which parents sought specialist consultation for their autistic child was reduced by 2 months for each additional parent-reported domain of atypical development in the first year of life. Stevens et al. (2016) conducted a study to compare the timing of parental concerns between autism and autism + ADHD groups. They concluded that both the first reported parental concern and the age at which a specialist assessment was sought were later for an autism + ADHD group of children than for those in an autism only group. There is also evidence that the nature of the concern that a parent presents to a specialist might aid in an earlier diagnosis of autism when the presenting concern fits with a symptom of the subsequent diagnosis, as the concern triggers that particular diagnostic pathway (Matheis et al., 2017; Rosenberg et al., 2011).

Once diagnosed, there is the question of whether parents feel that either or both diagnoses fit with the ongoing observed behaviour of their child. Researchers have argued for the co-occurring diagnosis to be further defined by the more dominant diagnosis, such as autism(+ADHD) or ADHD(+autism) (van der Meer et al., 2012). There is a gap in the literature pertaining to which diagnosis best fits with the ongoing behaviour that parents notice. One hypothesis is that parents of the children in the co-occurring group, might report that their children's observable behaviour is more aligned with the diagnosis more frequently given first; ADHD. This would fit with the hypothesis that the ADHD truly masks autism symptoms or is the more dominant observable behaviour (Davidovitch et al., 2015). In the ADHD only group and the autism only group, it is expected parents view the diagnosis as a more consistent summary of their child's behavioural symptoms.

The literature indicates that the timing, frequency and type of parent-reported atypical development and the nature and timing of specialist consultation might play a role in the age of an autism diagnosis (Zablotsky et al., 2017). Little is understood, however, about how patterns in these areas might compare across groups of autism only, ADHD only and a combined group. In particular an ADHD only group is often not included in comparative studies. This is a particularly important area of study given the hypothesis that children with both conditions resemble more closely early developmental ADHD patterns, rather than autism, which might account for the delay in autism diagnosis (Davidovitch et al., 2015; Soke et al., 2018).

This study aimed to build a clearer picture of why there might be differences in the timing of both autism and ADHD diagnosis when the two conditions co-occur, compared to when they are present in isolation. Specifically, a survey was conducted of

New Zealand parents of children with autism only, ADHD only, and autism + ADHD to examine the influence of various aspects of the diagnostic pathway on the timing of both autism and ADHD diagnosis across these three groups. These variables were: i) the number and type of parent-reported areas of atypical development in the first year of life; ii) the age at which parents felt specialist consultation was needed; iii) the behaviour that led to specialist consultation and whether this aligned with the child's subsequent diagnosis; iv) the wait time from when parents felt specialist consultation was needed to when a diagnosis was given; v) the types of specialist seen before a diagnosis; vi) any additional diagnoses; and vii) parental agreement that their child's behaviour matched their diagnosis.

Methods

Ethical Clearance and Informed Consent

Ethical approval for this study was provided by the Human Ethics Committee at Victoria University of Wellington, New Zealand [Approval number 28993]. Participation in the study was both voluntary and anonymous. Participants were asked not to include any information which would make them or their child identifiable. Participants were also informed that they did not have to answer any question and were given "prefer not to say" options.

Participants

Parents of children with autism, ADHD, or autism + ADHD were invited to participate in this survey. Participants were eligible for inclusion if they were: (i) parents, legal guardians, or caregivers (hereafter, parents), (ii) the child had a clinical diagnosis of autism, ADHD or autism + ADHD, reported by parents followed by a specified age of diagnosis, (iii) the child was aged 18 years or younger, and (iv) the

parents and child were living in New Zealand. Convenience sampling was used, as participants were recruited by an email sent by Autism New Zealand and ADHD New Zealand, as well as information about the study shared on various autism and ADHD social media groups. Participants were sent a link to the questionnaire within the advertisement.

Materials

The survey was hosted on Qualtrics from the 15th of March until the 1st of June, 2021. Many of the questions used in this survey were based on those in The Australian Autism Biobank's Family History Questionnaire (Alvares et al., 2018). The survey was revised after consultation with a researcher and advocacy advisor at Autism New Zealand. The questionnaire had 25 questions separated into 5 sections related to: (i) demographic characteristics, (ii) retrospective parent-reported atypical development, (iii) first concerns, (iv) specialists seen and additional diagnoses, and (v) age of diagnosis and fit of diagnosis from the parents' perspective.

The demographic characteristics section included questions about parent ethnicity, relationship to the child, highest education level, and total household income. Child demographic characteristics included gender, age, and the presence and birth position of siblings.

The atypical development section involved two questions. First, *"do you recall anything unusual about your child's development or behaviour during the first six months of his/her life?",* (referred to herein as parent-reported atypical development at ≤6 months) and "*Now think of the period between 7 and 12 months, can you recall anything unusual about your child's behaviour during that time?"* (referred to herein as parent-reported atypical development at 7-12 months). The parents then selected which

of 30 areas of atypical development were retrospectively relevant to their child at each age. These were based on the areas identified by (Waddington et al., 2022) and Guinchat et al. (2012). Some language was changed to be user-friendly, and categories that had no responses in (Waddington et al., 2022) were not included. Areas were identified by parents of autistic children as very early signs of atypical development, but were not necessarily specific to the autism criteria (Guinchat et al., 2012). The social development and stereotyped/restricted behaviour domains are associated with the diagnostic criteria for autism, and the hyperactivity and lack of attention subdomains within the temperament domain are associated with the diagnostic criteria for ADHD. The 30 areas were subdomains of seven domains of atypical development: language, social, stereotyped behaviour, motor, behaviour/temperament, medical, and abnormal physiological function. Parents could also select "other" and provide a description of any atypical development not covered in the list. These "other" atypical development descriptions were then coded in their entirety by WS (a researcher and an educational psychologist) and 20% were also independently coded by a research assistant. Domain and subdomain level agreement was calculated using the formula agreements/(agreements + disagreements) × 100, Domain-level agreement was 92.9 % at ≤6 months and 96.2 % at 7-12 months, sub-domain level agreement was 97.2 % and 98.0 % respectively.

The next section related to type and timing of the parental concern(s) that led them to seek specialist help. Parents were asked how old the child was when they first had concerns that warranted the need to see a specialist and to describe what particular concerns led to seeking specialist help in an open-text box. The concerns were then coded by WS (a researcher and an educational psychologist) according to whether the

concerns fit with a DSM 5 description of the particular diagnosis(es) the child obtained. The coding was based on ADHD concerns, autism concerns or behaviour/other category. A further 30% were coded by KC (a researcher and an educational psychologist) with an 88% interrater agreement.

The next section related to specialists seen, additional diagnoses and age of diagnosis. Parents were asked to select the types of specialists/professionals seen before receiving a diagnosis from a list and an "Other " option with text provided. The next question asked where their child was diagnosed with autism, ADHD or autism + ADHD. Depending on parents' responses, they were directed to the child's age of diagnosis for either autism only, ADHD only, or an age for both diagnoses. The last three questions of this section asked, "What diagnosis best described the behaviours of your child?" with the diagnostic options of autism and ADHD with main symptoms in brackets, a "both" and a "neither" option. A further two questions about any additional diagnoses with options including sensory processing disorder, epilepsy/seizures, oppositional defiant disorder, intellectual disability, global delay, learning disability and "other" and finally whether a misdiagnosis had occurred. A final section asked four open-ended qualitative questions, which are reported in the following publication: Willow J. Sainsbury, Chris J. Bowden, Kelly D. Carrasco, Andrew J. O. Whitehouse & Hannah Waddington (2023) Parent experiences of their children's diagnosis with autism, attention deficit hyperactivity disorder, or both conditions, *International Journal of Developmental Disabilities*, 1-11.

Data Analysis and Management

Data analysis was performed using IBM SPSS Statistics software (version 26). A chi-squared test of independence with Bonferroni correction determined whether

there were significant differences across diagnostic groups for gender, ethnicity, household income, highest education and absence or presence of siblings. A Kruskall Wallis test was used to analyse the age of participants across diagnostic groups.

Household income data was collapsed from 13 $NZ 10,000 increments to four $NZ 50,000-increments. In the highest level of education question many of the "Other" responses related to diplomas or trade qualifications, thus, a new category was added to encompass these responses. It was not possible to do an analysis for parent relationship to child due to having too few participants in any other category except mothers. Categories with fewer than 10 participants in each group were considered for exclusion or merging into a wider category to increase the reliability of group measures in statistical analysis. Excluded from statistical analysis of gender were 'non-binary' (n = 6) and 'prefer not to say' (n = 2) and excluded from analysis of siblings was the category of "twins" (n = 9). None of the "Other" ethnicities reached a threshold for analysis and were thus grouped together. Each participant's ethnicity was categorised as 'Māori' (the indigenous people of New Zealand) if one of their stated ethnicities was Māori.

Non-parametric statistical tests were also used to answer the research questions as the data was not normally distributed. Mann-Whitney-U tests were used to compare the age of autism and ADHD diagnosis, time to diagnosis, and the alignment of parents' description of behaviour with symptoms of the child's subsequent diagnosis. The autism + ADHD group was not included in the latter analysis due to the presenting behaviour description requiring symptom agreement of both an autism and ADHD diagnosis and most parents only described one presenting behaviour. Kruskall-Wallis tests were used to compare differences across the three diagnostic groups in terms of: (i) age of children at the time of the survey; (ii) total number of parent-reported atypical development; (iii)

age of from when specialist was sought; (iv) age when specialist was sought; (v) the number of different types of specialists seen, (vi) the total number of additional diagnoses across the three groups. The Kruskal-Wallis test presented the post-hoc p values in relation to each group combination and are reported. Chi-squared tests were used to compare differences across the three diagnostic groups in (i) atypical development in each domain at ≤ 6 and 7-12 months; (ii) alignment between parents' perceptions of the child's behaviour presented to a specialist and the child's subsequent diagnosis; (iii) alignment between diagnosis and parent perceptions of child's ongoing behaviour; and (iv) additional diagnoses. To control for multiple comparisons, a Bonferonni correction was used for post-hoc tests by multiplying all p-values by 3.

Results

Sample Characteristics

The parents of 355 children participated in the survey. Of these, 52 parents did not report their child's diagnosis, four parents reported that their children were over 18, and 11 only completed the demographic questions, and were thus excluded. This resulted in a final sample of 288 parents of children diagnosed with either (1) autism; (2) ADHD; or (3) autism + ADHD. Table 4.1 provides the demographic characteristics for these participants both overall and across diagnostic groups. In terms of the overall sample, the children were predominantly male with a mean age of 113 months (9.5 years). Mothers were more likely to have completed the survey. The parents who completed the survey were more commonly NZ European, had completed university and had a household income between $NZ 50,000 - 99,000. The average income in New Zealand is $NZ 67,000 (OECD, 2022).

Table 4.1.

Child and parent demographic characteristics (n = 288).

Demographic characteristic	Total n (%)/mean (months) (SD)	Autism only n (%)/mean (months) (SD)	ADHD only n (%)/mean (months) (SD)	Autism + ADHD n (%)/mean (months) (SD)	Missing data
Participant numbers	288	111	94	83	
Child gender					
Male	215 (74.7)	81 (73.0)	67 (71.3)	67 (80.7)	
Female	65 (22.6)	26 (23.4)	24 (25.5)	15 (18.1)	
Non-binary	6 (2.1)	3 (2.7)	2 (2.1)	1 (1.2)	
Prefer not to say	2 (0.7)	1 (0.3)	1 (0.3)	0 (0)	
Child age	113.4 (44)	97.9 (49)	118.3 (36)	128.7 (37)	
Age specialist needed		32.7 (27)	56.1 (33)	43.8 (26)	5
Child age when diagnosed with autism		63.1 (40)		87.6 (38)	0
Child age when diagnosised with ADHD			90.0 (28)	81.5 (26)	2

Parent relationship to child					
Biological mother	273 (94.8)	107 (96.4)	89 (94.7)	77 (92.8)	
Biological father	7 (2.4)	2 (1.8)	4 (4.3)	1 (1.4)	
Legal guardian/caregiver	8 (2.8)	2 (1.8)	1 (1.1)	5 (6.0)	
Siblings of child					
No siblings	31	9	13	9	
Younger siblings	149	58	55	36	
Older siblings	170	69	45	56	
Twins	9	4	2	3	
Parent ethnicity					1
NZ European (Pākehā)	200 (69.4)	70 (63.1)	70 (74.5)	60 (72.3)	
Māori	64 (22.2)	29 (26.1)	14 (14.9)	21 (25.3)	
Other: (Pacific People, Asian, European, South African, American, South American, Middle Eastern, African)	23 (8.0)	12 (10.8)	9 (9.6)	2 (2.4)	

Parent highest education				
< 12 years	36 (12.5)	14 (12.6)	14 (14.9)	8 (9.6)
12 years	75 (26.0)	34 (30.6)	20 (21.3)	21 (25.3)
Diploma/Trade	23 (8.0)	7 (6.3)	10 (10.6)	6 (7.2)
University	131 (42.4)	48 (43.2)	42 (44.6)	41 (49.4)
Prefer not to say/other (not specified)	23 (8.0)	8 (7.2)	8 (8.5)	7 (8.4)
Household income $NZ				
<$50,000	59 (20.5)	20 (18.0)	11 (11.7)	28 (33.7)
$50,000 - $99,999	105 (36.5)	44 (39.6)	39 (41.5)	22 (26.5)
$100,00-149,999	52 (18.1)	19 (17.1)	17 (19.1)	16 (19.3)
$150,000 or more	50 (17.4)	18 (16.2)	19 (20.2)	13 (15.7)
Prefer not to say	22 (7.6)	10 (9.9)	8 (8.6)	4 (4.8)

Note: Parent education and parent ethnicity relate to the parent who completed the survey. Pākehā is an indigenous term used for New Zealanders of European descent.

Chi-squared tests indicated that there were no significant differences across diagnostic groups in terms of highest level of education (X^2 (6, n = 265) = 4.40, p = 0.623), ethnicity (X^2 (4, n = 288) = 9.11 p=0.058) or gender (X^2 (2, n = 280) = 1.69, p = 0.429). A significant difference was found for the association between the diagnostic group and having a younger sibling (X^2 (2, n = 288) = 7.73, p=0.021) with post-hoc testing indicating a lower proportion of younger siblings in the ADHD group compared to the autism or co-occurring group. There were no other significant differences in the likelihood of having no siblings (X^2 (2, n = 288) = 1.74, p=0.420) or older siblings (X^2 (2, n = 288) =4.06, p = 0.131) across diagnostic groups. There were significant differences in family income across diagnostic groups (X^2 (6, n = 266) = 14.66, p = 0.023). Post-hoc testing indicated that the autism + ADHD group had a smaller proportion of participants in the $NZ50,000-99,999 group compared to both the autism and ADHD only groups.

A Kruskal-Wallis test determined that there were significant differences in child age across diagnostic groups (H(2) = 27.331, p < .001). Post-hoc testing determined that the age of the autism group was significantly lower compared to those with both autism + ADHD and those diagnosed with ADHD, p <.001 and p = 0.045, respectively. The age of ADHD children was also significantly younger than the autism + ADHD group (p = .040).

Age of Diagnosis

Mann-Whitney U tests indicated that an ADHD diagnosis was given significantly earlier for an autism + ADHD (Mdn = 79.0) (months) group than for an ADHD only group (Mdn = 96.2), U (n = 83, n = 92) = 3067.00, z = -2.25, p = 0.025, while autism diagnosis was given significantly earlier for an autism only (M = 81.0) than for an autism + ADHD (Mdn = 119.6) group, U (n = 111, n = 83) = 2771.50, z = -4.75, p< 0.001.

Atypical Development at ≤ 6 Months

The percentage of participants reporting atypical development in each domain and subdomain across diagnostic groups is reported in Table 4.2.

Table 4.2.

Percentage of parents reporting atypical development in each domain and subdomain across diagnostic groups when their children were ≤ 6 and 7-12 months (n = 288).

Domain-/Subdomain	≤ 6 months			7-12 months		
	Autism only	ADHD only	Autism+ ADHD	Autism only	ADHD only	Autism+ ADHD
Reported at least one area of atypical development across domains	59.5	54.3	60.2	82.0**	73.4**	86.9**
Language	**23.4**	**12.8**	**24.1**	**53.2****	**19.1****	**43.4****
Delayed speech/vocalizations	17.1	9.6	16.9	47.7	18.1	37.3
No speech/vocalizations	11.7	4.3	9.6	13.5	4.3	9.6
Poor language comprehension	15.3	7.4	10.8	33.3	8.5	21.7
Loss of language	2.7	0.0	3.6	15.3	2.1	10.8
Social development	**30.6**	**17.0**	**22.9**	**59.5****	**23.4****	**45.8****
Delayed social communication	15.3	8.5	12.0	37.8	10.6	26.5
No social communication	8.1	2.1	6.0	21.6	1.1	10.8
Gaze abnormalities	18.0	7.4	13.2	34.2	10.6	31.3
Poor social interaction	15.3	10.6	14.5	38.7	17.0	31.3
Lack of response to social stim.	21.6	5.3	10.8	41.4	8.5	26.5
Stereotyped/restricted behavior	**38.7**	**30.9**	**34.9**	**64.0***	**47.9***	**69.9***
Stereotyped movements	20.7	5.3	12.0	41.4	12.8	24.1
Need for routine/rituals	21.6	14.9	20.5	43.2	26.6	38.6
Stereotyped/restricted interests	9.9	3.2	10.8	40.5	11.7	32.5

Preoccupation with object	14.4	7.4	10.8	40.5	19.1	28.9
Hypersensitivity	25.2	14.9	21.2	48.6	23.4	51.8
Hyposensitivity	9.0	2.1	4.8	18.0	5.3	13.3
Motor development	**31.5**	**20.2**	**26.5**	**45.9***	**25.5***	**41.0***
Motor delay	20.7	8.5	16.9	37.8	20.2	31.3
Hypotonia	6.3	3.2	10.8	9.0	4.3	15.7
Hypertonia	4.5	2.1	2.4	8.1	2.1	3.6
Swallowing/Sucking	14.4	13.8	19.3	11.7	7.4	13.3
Behavior/Temperament	**37.8**	**39.4**	**44.6**	**56.8**	**57.4**	**71.1**
Lack of attention	7.2	10.6	14.5	21.6	18.1	34.9
Hyperactivity	6.3	20.2	16.9	20.7	33.0	43.4
Passivity	9.0	4.3	9.6	9.0	4.3	10.8
Tantrums/opposition	11.7	7.4	12.0	36.9	22.3	39.8
Difficult to soothe	26.1	24.5	33.7	32.4	27.7	39.8
Aggression/violence	3.6	2.1	3.6	10.8	2.1	14.5
Self-harm	3.6	0.0	1.2	10.8	1.1	4.8
Extreme attachment to caregiver	20.7	10.6	19.3	30.6	20.2	37.3
Medical issues	**12.6**	**19.1**	**10.8**	**15.3**	**19.1**	**15.7**
Another condition	1.8	4.3	1.2	0.9	5.3	1.2
Sickness	11.7	17.0	9.6	14.4	14.9	14.5
Abnormal physiological function	**34.2**	**36.2**	**42.2**	**56.8**	**48.9**	**50.6**
Sleeping	27.0	35.1	39.8	41.4	45.7	49.4
Feeding	18.0	11.7	18.1	35.1	13.8	20.5

Note: Items in bold are domains while indented, non-bolded items are subdomains and asterisks indicate p value (*p = 0.05 **p = 0.001).

A Kruskal-Wallis test was used to compare total number of domains of atypical development indicated by parents and there was no statistically significant difference between groups in atypical development ($H(2) = 1.119$, $p = 0.572$). Chi-square test for the presence or absence of any area of atypical development did not significantly differ between the three groups, (X^2 (2, n = 288) = 809, p = 0.667.

Chi-square tests revealed no significant difference across diagnostic groups in domains of language (X^2 (2, n =288) = 4.73, p = 0.94); social (X^2 (2, n = 288) = 5.25, p = 0.73); stereotyped behaviour (X^2 (2, n =288) = 1.39, p = 0.50); motor (X^2 (2, n = 288) = 3.36, p = 0.19), behaviour (X^2 (2, n = 288) = 0.95, p = 0.62); medical issues X^2 (2, n = 288) = 2,88, p = 0.24); or abnormal function (X^2 (2, n = 288) = 1.34, p = 0.51).

Atypical Development at 7-12 Months

A Kruskal-Wallis test was used to compare total number of domains of atypical development indicated by parents and there were significant differences between groups ($H(2) = 16.149$, $p < .001$). Bonferroni-adjusted pairwise comparisons determined that ADHD had significantly fewer parent-reported areas of atypical development than autism ($p<0.001$) and autism + ADHD ($p = 0.014$) with no significant difference between autism and autism + ADHD (p=1.000). Chi-square test for the presence or absence of any area of atypical development did not significantly differ between the three groups, (X^2 (2, n = 288) = 5.25, p = 0.073).

There were no significant differences in domains of behaviour (X^2 (2, n = 288) =4.89, p = 0.087), medical issues (X^2 (2, n = 288) =.62, p = 0.732) and abnormal function (X^2 (2, n = 288) = 2.50 p = 0.287). There were significant differences in the language domain (X^2 (2, n = 288) =225.52, $p < 0.001$). There were a higher proportion of parents reporting atypical development in the language domain if their child had autism than autism + ADHD, and a significantly lower proportion for the ADHD group compared to both autism and autism +

ADHD groups. There were also significant differences across diagnostic groups in the social domain (X^2 (2, n = 288) = 27.08, p <0.001), with the a higher proportion in the autism group reporting atypical social development than the ADHD and autism + ADHD groups and a lower proportion in the ADHD group than all other groups.

There were significant differences in the stereotyped behaviour domain (X^2 (2, n = 288) = 9.88, p = 0.007), with a greater proportion of parents in the autism + ADHD group reporting atypical stereotyped behaviour than the ADHD group, and the ADHD group had a lower proportion than the autism and the autism + ADHD groups. There were also significant differences in the motor domain (X^2 (2, n = 288) =9.95, p = 0.009) with a higher proportion of parents reporting atypical motor development in the autism group and a lower proportion of parents reporting atypical development in the ADHD group.

Age Specialist Needed

A Kruskal-Wallis H test indicated that the age (months) at which parents felt specialist help was needed differed between the three diagnostic groups (H(2) = 36.647, p <.001). Post-hoc analysis determined that the age at which a specialist was needed was significantly lower in those with autism compared to those with both autism + ADHD and those diagnosed with ADHD alone, (H(2) = 17.040 , p <.001) and (H(2) = 35.759, p <.001) respectively. The age a specialist was needed did not differ between the autism + ADHD and ADHD groups (H(2) = 2.290, p = .391).

Further analysis of the specific behaviours which parents report led them to seek specialist help are shown in Table 4.3. The behaviours parents presented were coded for whether they aligned with the child's subsequent diagnosis. The differences in the agreement of type of presenting behaviour of the subsequent diagnosis was significant X^2 (2, n = 288) = 96.45, p < 0.001. Parents of children with autism most frequently presented with autistic

behaviour(s). The ADHD and the autism + ADHD group showed far greater inconsistency with a greater reporting of behaviours that do not fit with an autism or an ADHD diagnosis.

Table 4.3.

Alignment between behaviours that led parents to seek specialist help and diagnosis

Diagnostic Groups	Behaviours related to Autism only (n/%)	Behaviours related to ADHD only (n/%)	Behaviours related to Autism +ADHD (n/%)	"Other" behaviours
Autism	91 82%	0 0%	0 0%	20 18%
ADHD	16 17%	39 41.5%	1 1.1%	38 40.4%
Autism + ADHD	32 38.6%	5 6%	11 13.3%	35 42.2%

Wait Time from Age Specialist Needed to Age of Diagnosis

Mann-Whitney U tests were used to examine the time (months) between when a specialist was needed to diagnosis for ADHD and autism groups compared to the autism + ADHD group. Analysis indicated that the wait time from age parents felt a specialist was needed to diagnosis for ADHD only group and the co-occurring group was not significant U (n = 162) = 3 506.00, z = 0.763 p = 0.445. The wait time from specialist contact to diagnosis for the autism only and autism + ADHD group were significant with the autism only group (M = 82.83) waiting significantly less time for an autism diagnosis than an autism + ADHD group (M = 109.53), U(n = 188) = 5582.50, z = 3.345 , p = 0.001.

Alignment of parents' description of behaviour with the child's subsequent diagnosis and the effect on time to diagnosis.

A further Mann-Whitney U analysis examined whether the type of behaviour described by parents as motivating specialist help aligned with a symptom of the child's subsequent diagnosis and whether this affected time to diagnosis. In the autism only group, parents who described a symptom of autism as motivating them to seek specialist help waited significantly less time for a diagnosis than parents of autism only children who presented with concerns that did not indicate a symptom of autism, U (n = 91, n = 20) = 616.50, z = -2.254, p = 0.024. The same was true within the ADHD only group. Parents who presented with concerns that indicated a symptom of ADHD waited significantly less time than parents who presented with concerns that did not indicate a symptom of ADHD, U (n = 37, n = 55) = 444.00, z = -4.572 p < 0.001.

Number of types of specialists seen

A Kruskal-Wallis test was used to compare the number of different types of specialists seen between the three diagnostic groups, determining that at least one group differed ($H(2) = 14.776$, p = 0.001). Bonferroni-adjusted pairwise comparisons determined that the number of types of specialists seen was significantly higher in those with autism + ADHD compared to those diagnosed with ADHD alone and autism alone (p < .001 and p = .044, respectively). There was no difference in the number of types of specialists seen between the ADHD and autism only groups (p = 0.340).

Total additional diagnoses

A Kruskal-Wallis test was used to compare the number of additional diagnoses between the three diagnostic groups, determining that no group significantly differed; H(2) = 4.86, p =.089. However, certain diagnoses had a significant correlation with particular diagnostic groups. Oppositional defiance disorder was more likely to be diagnosed in

addition to ADHD than autism and autism + ADHD (X^2 (2, n=288) =26.55 p =< 0.001) and sensory processing disorder was more likely to be diagnosed with autism than ADHD and autism + ADHD (X^2 (2, n=288) =13.21 p < 0.001). Intellectual disability (ID) was not significant across the groups (n = 9 ADHD only; n = 15 Autism; n = 15 Autism + ADHD); (X^2 (2, n=288) =2.72 p = .257

Diagnosis and parent perceptions of child's behaviour

There were no significant differences between groups about whether parents believed that the diagnosis explained their child's behaviour (X^2 (2, n = 288) =1.75 p = 0.417). Within the ADHD group, 25% of parents disagreed that ADHD was the best diagnostic description for their child's behaviour and all selected both autism + ADHD as a better explanation. Within the autism group, 20.7% disagreed that autism was the best diagnostic description for their child's behaviour and selected both except for 3 parents, who opted for neither. Within the co-occurring group, 16.9% disagreed that autism + ADHD was the best diagnostic description for their child's behaviour and selected either ADHD (n = 8) or autism (n =7).

Discussion

This study sought to understand similarities and differences in early childhood diagnostic experiences for children with diagnoses of autism, ADHD or both. In line with previous research, ADHD diagnoses were significantly earlier for the autism + ADHD group compared to the ADHD only group, and autism diagnoses were significantly later for the autism + ADHD group compared to the autism only group (Sainsbury et al., 2022). The results provide further data that children with autism + ADHD may present with a different and more complex diagnostic picture. This presentation of retrospective parent-reported atypical development appeared as a "half-way" point with higher reported atypical development than an ADHD group, but lower than an autism group. Along the diagnostic pathway the co-occurring group resembles more closely the characteristics of an ADHD

group, such as with timing of when a specialist was needed. In other measures, an autism + ADHD group presented with neither autism nor ADHD patterns such as parents' descriptions of the concern that led to specialist help being more ambiguous, possibly contributing to the co-occurring group having longer wait times and seeing more types of specialists between seeking help to diagnosis than the autism only or ADHD only groups.

The parent reported observation of atypical development demonstrated that under 6 months there were no significant differences across diagnostic groups, but by 12 months statistically significant differences were occurring. The significant differences between increased atypical development reported in the autism only group compared to the ADHD group, was to be expected. Core autism behavioural features of language, stereotyped behaviour and social atypical development were significantly more reported in the autism group than the ADHD which fits with diagnostic models which suggest the ADHD symptoms are more apparent in later childhood and autism symptoms are observable at 12 months (Ozonoff et al., 2014). In Wallisch et al. (2020)'s study of a parent population who had 3-5 year olds diagnosed with multiple diagnoses, they found similar patterns of atypical parent reported concerns with language, and social atypical development was reported less in an ADHD group than an autism group. Early signs of ADHD are usually still considered to occur during toddler years, however, recent research is finding that, like autism, atypical development is seen in the first year of life with feeding and sleeping difficulties and motor difficulties, in children subsequently diagnosed with ADHD (Athanasiadou et al., 2020). Parents of children with ADHD did report sleeping difficulties at similar rates to the other diagnostic groups.

The autism + ADHD diagnostic group had fewer areas of reported atypical development than the autism group but more than the ADHD group. The prevalence of atypical language and social development in the first year of life appeared to be markedly less

for autism + ADHD group. Atypical language development is a known trigger for early help-seeking and less atypical development in this area is likely to contribute to a delay in an autism diagnosis (Zablotsky et al., 2017). However, this trend was reversed with stereotyped behaviour, whereby parents of autism + ADHD children reported higher atypical development in this area than autism and ADHD children. This might be a possible focus for diagnostic identification in children with co-occurring autism and ADHD.

The significant difference in the average age when parents first sought a specialist differed between the three different groups, suggesting the delay in diagnosis may start, in part, with a delay in parents of children with autism + ADHD seeking specialist help compared to an autism only group (Stevens et al., 2016). The age at which parents of the co-occurring group sought specialist help was significantly later than the autism only group, but not statistically different from the ADHD group. In this way the co-occurring group fits an ADHD pattern. The delay in help seeking for these groups might be due, in part, to the nature of the child behaviour in that might be attributed to temperament, behavioural difficulties or parenting rather than requiring specialist intervention (Wallisch et al., 2020; Yamauchi et al., 2015).

Parents presenting with a specific symptom-related behaviour which corresponds directly to a particular diagnosis are likely to trigger a particular diagnostic pathway (Becerra-Culqui et al., 2018). The autism group was the most consistent in terms of presenting behaviours that described a symptom of autism. Analysis demonstrated that when parents presented behaviour that described a symptom of a particular diagnosis in either the ADHD or the autism group, those families had a shorter wait time to diagnosis than parents who presented with a less diagnostic-specific behaviour.

Parents of the co-occurring group described presenting behaviours to specialists which did not fit with key symptoms that might trigger a diagnosis of either condition, but

rather the most common concerns parents described appear to be "other", including behaviour, then autism and then ADHD symptoms. This might result in a less clear referral to a specialist or a referral to a specialist who does not have a specialty in autism or ADHD diagnosis. Evidence of this was seen in the increased number of types of specialists that the co-occurring group saw before receiving a diagnosis. Eggleston et al. (2019)'s study of parents' experiences of their child receiving an autism diagnosis found that co-occurring ADHD was also predictive of a higher number of professionals consulted prior to diagnosis, (Eggleston et al., 2019). It is also interesting that there were a lower number of parent-described ADHD behavioural symptoms that initiated specialist help in the co-occurring group, given that an ADHD diagnosis is often made earlier compared to an ADHD only group, and ADHD is more commonly diagnosed first in a co-occurring group (Kentrou et al., 2019). Thus, it was hypothesized that parents would describe behaviour that was more likely to flag ADHD symptoms. However, in some ways this group did resemble the ADHD group because of the high number of "other" behaviours. Primarily behavioural concerns have been shown to be associated with delays in seeking specialist help, possibly due to parents thinking the behaviours are due to the temperament of the child or perceived as a parenting issue (Ghanizadeh, 2007; Yamauchi et al., 2015).

This study elucidated further where along the diagnostic pathway delays in diagnosis might be occurring. Stakeholders in the diagnosis and treatment of children might consider that parents of children with autism + ADHD often present with later behavioural and other types of concerns, and with less parent-reported atypical development of language and communication than an autism only group. A further consideration might also be that parents presenting with concerns around behaviour and unclear referrals may warrant a more direct referral to a multidisciplinary team rather than referrals to a number of types of individual practitioners. Referrals that trigger specific developmental symptoms of a particular diagnosis

are likely to encounter a more straightforward diagnosis pathway. In addition, specialists might consider that a first diagnosis of ADHD should not preclude further assessment of autism, or that autism should at least not be ruled out. Specialist might also want to consider discussing with parents, whose children are first diagnosed with ADHD, the key symptoms of autism so that parents feel empowered to return for the possibility of an autism diagnosis sooner. These suggestions have the potential to reduce the delay in diagnosis for autism + ADHD children and improve the diagnosis and treatment for children and their families.

One limitation of this study is that it did not use an epidemiological sample, and may not be representative of the broader population of these groups. A more educated and homogenous sample of society often answer online surveys (Eggleston et al., 2019). However, the mean averages of the survey represented a cross section of New Zealand based on the average highest education and income of the sample (OECD, 2019). In terms of ethnicity, Māori, the indigenous population of New Zealand, are sufficiently represented in this sample (Tupou et al., 2021).

The categories of diagnosed children representing autism, ADHD and autism + ADHD are not static, and it is possible that children in the single diagnosis categories will, at some point in the future, move into the co-occurring group. However, this possible movement does not detract from the snapshot in time of these diagnoses and how they might relate to atypical development, timing and type of parental concerns, diagnosis experience and current diagnosis of the child. To enhance the level of evidence further studies might also look at the risks of mis-diagnosing co-occurring autism + ADHD. A greater understanding of any undesirable consequences in giving a younger child a co-occurring diagnosis should be addressed.

There are limitations for parents self-reporting. The first concerns that led to specialist help also has a recall bias, particularly once diagnosis is known, and parents might

retrospectively recall concerns that fit with the diagnosis. The survey uses parental report as the basis for diagnostic criteria, which had not been clinically confirmed. Miodovnik et al., (2015), however, cite research in both autism and ADHD areas that found convergent validity between parent reported ADHD diagnosis data from insurance claims and autism diagnosis data form nationally representative surveys. In addition, data about first concerns and diagnostic history were collected at one point in time for children whose ages ranged from 2 to 18 years of age. All parents were asked to reflect over time, and parents of the oldest children needed to reflect over the longest period which might create the possibility of differential recall biases as it relates to the parents' report, particularly for the co-occurring diagnostic category with, on average, older children than the autism or ADHD group. A further consideration is differentiating between early atypical concerns only 6 months apart which is retrospectively remembered, and therefore, the data might not accurately reflect the progression of atypical development in this early year of life.

Conclusion

The study found that the diagnosis of autism in an autism + ADHD population is delayed. A delayed diagnosis is significant for this high need population as it reduces the opportunities for early intervention. The delay in diagnosis may be due to parents noticing less atypical development in language and social behaviours, the presentation of less autism-specific concerns to a specialist and at a significantly later time than an autism only group. A lack of flagging for a diagnosis potentially might result in multiple referrals to different supports and specialists and possibly a longer diagnostic pathway. These factors may contribute to a delay in an autism diagnosis for an autism + ADHD child.

CHAPTER 5

STUDY 2: Parent experiences of diagnosis for children with autism, ADHD, or both conditions

Chapter Note

This chapter was published as a journal article in the *International Journal of Developmental Disabilities,* titled *Parent experiences of their children's diagnosis with autism, attention deficit hyperactivity disorder*. Dr Hannah Waddington helped with the design, analysis and editing. Dr Chris Bowden guided on the use of reflexive thematic analysis. Professor Andrew Whitehouse and Dr Kelly Carrasco assisted with editing. I designed, conducted and wrote this article and it is entirely my work aside from the input specified above. Aside from changes to ensure consistency of formatting with the rest of the thesis, this chapter is identical to the published article. The complete citation for the published article is: Sainsbury, W.J., Bowden, C.J., Carrasco, K.D., Whitehouse, A.J.O., Waddington, H. (2023) Parent Experiences of their Children's Diagnosis with Autism, Attention Deficit Hyperactivity Disorder, or Both Conditions. *International Journal of Developmental Disabilities*, 1 -11. https://doi.org/10.1080/20473869.2023.2166197

Abstract

Purpose: A comparison of parents' experiences of getting a diagnosis for their child with autism, ADHD and both diagnoses can inform our understanding of common and unique themes across these neurodevelopmental conditions. **Method**: A quantitative and qualitative online anonymous survey of 288 New Zealand parents of children diagnosed with autism (n = 111), ADHD (n = 93), or both conditions (n = 84) was conducted. Open-ended questions were analysed using reflexive thematic analysis. **Results:** Parents described an adversarial diagnosis and support system where seemingly arbitrary criteria and thresholds were applied. Key themes specific to the different diagnostic groups were also identified. **Conclusions**:

Common themes of parents' experience across the different neurodevelopmental conditions highlight the need for changes to the diagnostic process.

Key words

Autism, ADHD, co-occurring, diagnosis, experience, reflexive thematic analysis.

Autism spectrum disorder (autism) and attention-deficit/hyperactivity disorder (ADHD) are neurodevelopmental conditions and typically emerge in early childhood (American Psychiatric Association, 2013). The conditions are separated by core symptoms; the autism diagnostic criteria include fixed, rigid behaviour and social difficulties, and ADHD is characterised by the presentation of attention difficulties, impulsivity and hyperactivity (American Psychiatric Association, 2013). Prevalence estimates in children are 5% for ADHD (Sayal et al., 2018) and 1.85% for autism (Maenner et al., 2020). The conditions also have high rates of co-occurrence with an estimated 32% of autistic 8 year-olds also having an ADHD clinical diagnosis (hereafter referred to as autism + ADHD) (Soke et al., 2018).

Obtaining a diagnosis in childhood for autism or ADHD often follows a typical diagnostic pathway. It begins with parents noticing their child’s atypical development, they then present with a particular concern(s) to a health professional before being referred to a specialist or a multidisciplinary team for diagnosis (Gibbs et al., 2019; Sayal et al., 2018). In New Zealand, parenting courses (e.g., Incredible Years Parent Programme®) are often offered by agencies and professionals before a referral to specialist as a means to rule out behaviour due to challenges around parenting (Fergusson et al., 2009). The autism diagnostic process will have cultural differences across national contexts, but is described in qualitative analyses in United Kingdom and Europe as remarkably similar to a New Zealand context (Braiden et al., 2010; Legg & Tickle, 2019), which has led to direct comparisons across

national borders of the process (Eggleston et al., 2019). Fifty-six percent of parents in the United Kingdom (Crane et al., 2016) and 37% of parents in New Zealand (Eggleston et al., 2019) reported finding the autism diagnosis process very stressful. Similarly, across 10 European countries 33% of parents reported ‘a great deal of difficulty’ obtaining a referral and 31% ‘a great deal of difficulty’ obtaining a diagnosis of ADHD (Fridman et al., 2017).

Barriers to getting a diagnosis have been identified in large scale quantitative surveys (Crane et al., 2016; Eggleston et al., 2019; Fridman et al., 2017) and qualitative thematic analyses (Boshoff et al., 2021; Elder et al., 2016; Makino et al., 2021; Ryan & Salisbury, 2012). Barriers to diagnosis for both autism and ADHD include: the delay caused by seeing multiple specialists (Eggleston et al., 2019; Fridman et al., 2017), lengthy wait periods between presenting with a concern and eventual diagnosis (Fridman et al., 2017; Stevens et al., 2016) and difficulty accessing specialists who can diagnose reliably (Crane et al., 2016; Fridman et al., 2017). Parental distress can be compounded by premature reassurances that their children are developing typically, or 'therapeutic nihilism' whereby they are told nothing can be done so the diagnostic pathway is ignored (Elder et al., 2016; Ryan & Salisbury, 2012).

The factors that affect stress and create barriers during the diagnostic process often delay diagnosis and thus the opportunity for early intervention. Early intervention has been shown to be effective, with greater adaptability and life outcomes for children who obtain early evidenced-based interventions, particularly for autism (Whitehouse et al., 2021), but the evidence for the effectiveness of early intervention for ADHD is also mounting (Bannett et al., 2022). However, recent research on the wider effects of ADHD diagnosis indicates there are concerns around early diagnosis and some issues related to the impact of a possible diagnostic shift.

Recent studies compared matched symptomatic young people diagnosed with ADHD and without, and found that young people with the label showed the same or worse on quality of life and relationship measures than those without the label (Kazda et al., 2022; O'Connor & McNicholas, 2020). O'Connor et al. (2018) have highlighted the instability of childhood diagnoses and the ramifications for young people and their families when diagnostic shift occurs, that is, when a diagnosis is retracted, replaced, or supplemented. Clinicians can experience dilemmas when weighing up providing an early but possibly inaccurate diagnosis versus providing early access to support (Mitchell & Holdt, 2014). Diagnostic labels are also tools by which parents and young people make sense of their behaviour, their identity and stigma, therefore, it is important that accurate diagnosis occurs (O'Connor et al., 2018).

The most uncommon diagnostic shift and the one that has the most negative impact, is diagnostic retraction, so O'Connor et al. (2018) speculate that the easiest option is to let a previous diagnosis 'recede from active clinical attention' (pg. 989), rather than retraction. However, the aim of this study is not to explore difficulties with accuracy of diagnosis, the system by which families can access support or whether they should begin the journey, but to examine the diagnostic process itself from the parent's perspective. It has been shown that high satisfaction with the diagnostic process leads to lower levels of stress, better coping strategies, and quicker adoption of evidence-based interventions by parents (Crane et al., 2016). Therefore, improving understanding of and the experience of the diagnostic process is essential to improved immediate outcomes for families, who have already begun this process.

Previous studies have explored the issues and challenges that parents face getting a diagnosis and support for children with autism and, to a lesser extent, ADHD (Carr-Fanning & Mc Guckin, 2018; Makino et al., 2021). Qualitative analyses and systematic reviews have used several frameworks and identified key themes that give greater context to the parents experience of the autism diagnostic process (Legg & Tickle, 2019). Legg and Tickle (2019)

identified three key types of parent need; emotional, relational, and informational. These needs change over the diagnosis process. Parental emotional guilt during the search for a diagnosis was replaced by grief or affirmation of autism identity post diagnosis (Legg & Tickle, 2019). Information needs also changed with parents requiring more information in different formats at different points highlighting the need for strong relationships between clinicians and family (Legg & Tickle, 2019). The review also showed some parents of children with autism in the UK experienced dismissal of their concerns, longer wait times, and unsatisfactory assessment, communication of diagnosis and supports which contradicted best practice guidelines (Legg & Tickle, 2019).

In addition, qualitative analyses and systematic reviews show several frameworks have been used in research to understand: (i) how parents cope with an autistic label for their child (O'Connor et al., 2018), (ii) causal-blame attribution (Dale et al., 2006), (iii) grief or autistic identity celebration and (iv) whether they adopt a social or medical model of their child's needs (Russell & Norwich, 2012). However, in all cases parents appear to have complex and evolving understandings. Russell and Norwich (2012) identified that parents teeter between wanting to normalise their child's status and resist diagnosis and the opposite of wanting to move towards diagnosis and advocating for the normalisation of autism.

However, no study has qualitatively compared the experience of getting a diagnosis across autism, ADHD and both diagnoses. The increasingly prevalence of co-occurring neurodevelopmental conditions (Soke et al., 2018) means that understanding transdiagnostic experiences will inform an understanding of the diagnosis process. Autism + ADHD are one of the most experienced co-occurring coupling, therefore comparative insights of a co-occurring group should strengthen and provide a more complex and nuanced understanding of the diagnostic process and experience. The seemingly different diagnosis pathways should suggest markedly different experiences because an autism diagnosis is possible from 12

months (Pierce et al., 2019) and usually requires an interactive-observational schedule (Hong et al., 2020), an ADHD assessment requires a multiple setting rating scale questionnaire and usually occurs after the age of 6 years (Izzo et al., 2019). However, any similarities in the experience, which sit outside of the expected differing developmental milestones or assessment tools, could suggest ecological and contextual factors that shape the process itself.

The current online survey aimed to collect a qualitative data to understand parents' experiences and perceptions of the diagnosis process for either autism, ADHD or autism + ADHD. The parents' perceptions reported in this article were drawn from open-ended questions (qualitative), which were part of a broader survey that also included closed questions (quantitative) examining the diagnostic pathway for these three groups in New Zealand. The value of open survey questions as a research method is that this is a 'wide-angle lens' that can include a number of diverse voices, wide geographical participation, encourage openness in responses, and provide participants with autonomy and control over their participation (Braun et al., 2020). Reflexive thematic analysis was used to analyse the qualitative data due to the extensive response rate and depth of answer that required an understanding of latent themes, not anticipated at the outset (Braun & Clarke, 2021). This analysis was designed to aid our understanding of the diagnostic experience across these three groups.

Methods

Ethical Clearance and Informed Consent

Ethical approval for this study was provided by the Human Ethics Committee at Victoria University of Wellington, New Zealand [Approval number 28993]. Participation in the study was both voluntary and anonymous. All participants provided written and informed consent prior to enrolment in the study.

Participants

Parents of children with a diagnosis of autism, ADHD, or co-occurring autism and ADHD were invited to participate in a survey, eligibility criteria being: (i) they were parents, legal guardians, or caregivers (hereafter, parents), (ii) their child had a clinical diagnosis of autism, ADHD or autism + ADHD, (iii) their child was aged 18 years or younger, and (iv) parents and child were living in New Zealand. A purposive, criterion-based convenience sampling strategy (Palinkas et al., 2015) was used to recruit a diverse range of participants who had children diagnosed with the three conditions. Participants were recruited by an email sent by Autism New Zealand and ADHD New Zealand and information about the study shared on various autism and ADHD social media groups. Participants were sent a link to the questionnaire within the advertisement.

Materials

This paper analyses qualitative data from an anonymous online survey hosted on Qualtrics (data collected from 15th March 2021 to 1st June 2021) on the early development, diagnosis and experience of obtaining a diagnosis for parents of children, diagnosed with autism, ADHD or both in New Zealand. This was a quantitative and qualitative design (Braun et al., 2020). The survey included a combination of 25 closed (quantitative) questions and open-ended (qualitative) questions separated into six sections: (i) demographic characteristics, (ii) atypical development, (iii) first concerns, (iv) specialists seen, additional diagnoses, and (v) age of diagnosis and (vi) qualitative questions. The quantitative questions in sections (i-v) were used to obtain a comprehensive picture of characteristics of participants, their children, key aspects of diagnostic experiences and types of specialists seen. The details and analysis of sections (ii) - (v) are reported in the quantitative analysis (Sainsbury et al., 2022). The open-ended (qualitative) questions in section (vi) focused on parent's experiences of the diagnosis pathway and were designed to be broad, and to elicit

both strengths and weaknesses of the diagnosis process, while also giving participants an opportunity to share other information about the diagnosis process. The three qualitative questions were: (1) What was helpful during the diagnostic process? (2) What was unhelpful about the diagnostic process? (3) What else would you like researchers to know about your experience of getting a diagnosis for your child? The survey questions and design were informed by consultation with a research and advocacy advisor at Autism New Zealand.

Data Analysis

Descriptive statistics were used for demographic data and to describe the response rate and quantity across the quantitative responses in the survey. Reflexive Thematic Analysis (RTA) is a qualitative method emphasising the subjectivity of the researcher as a resource to be utilised in a continual reflexive engagement at all stages of the thematic process (Braun & Clarke, 2021). Unlike other qualitative methodologies it is not tied to a particular epistemological or theoretical perspective (Maguire & Delahunt, 2017), however, Braun and Clarke (2021) articulate that reflexive thematic analysis is not done in a ‘vacuum’ and researchers must consider the standpoint from which they analyse (Braun & Clarke, 2021). A prerequisite for reflexive thematic methodology was a rich data set of qualitative responses. The high rate of participation and length of responses suggested this approach would be an appropriate method to analyse parents' survey responses.

Braun and Clarke (2021) reflexive TA has six key steps: The first and the second step involved reading and re-reading the data as a whole to obtain an initial impression of the experience and meaning and using an inductive approach to generate initial codes. The researcher coded participant responses by hand using open-coding, not using any pre-set codes but developing and modifying codes as she went through the process. This led to the third stage of reflexive TA and the creation of semantic (surface descriptive level) codes based on participants' language, such as 'child labelled naughty,' 'reluctance to diagnose'.

These codes were then interrogated and refined again (recursive analysis). Participants' responses were also placed along a diagnostic pathway, such as ordering responses into initial concerns, encounters with professionals, and post diagnosis support.

This led to the third step construction of topics corresponding to a diagnosis pathway and to codes suggestive of barriers and enablers at each step. All codes were grouped and corresponding quotes were listed under each to test the value, to describe the data and as an indication of prevalence to ensure key patterns across the dataset were being captured (Braun et al., 2020). The quotes may have been changed for spelling clarification and are not necessarily the full response of the participant (Terry et al., 2018).

The fourth step involved the search for themes and patterns across the data. This involved ‘dwelling with’ data and multiple reiterations of the data (Braun & Clarke, 2021). The guiding questions were: ‘Has the meaning behind the participant's comments been captured? What has been left out and why?’ Reflexivity was enhanced through the use of a self-reflective journal and visual modelling which was used to describe decisions and coding (e.g., development, revision, expanding a collapsing of codes and themes). Supervision and critical discussion of data, codes, construction of themes with peers also occurred (Braun and Clarke, 2021). This was structured by the authors' (C.J. Bowden) and (H.Waddington) reviewing codes and themes and challenging greater reflexivity in the data.

This led to the latent analysis and a deeper interpretation and consideration of meaning of the experience for parents in their journey for diagnosis and support.

The fifth step involved defining and naming themes and returning to steps two and three to check accuracy and meaning of initial coding attempts, and mapping where the themes interacted and the relationships between themes. The coherency, overlap and distinctiveness of themes was also scrutinised with the acknowledgement that some subthemes were shared between themes. During this phase the specificity of the child's

diagnosis became relevant to seeing where themes were represented by all diagnostic categories, or when only one or two diagnoses were indicated in the analyses. The final step was writing up the analysis, research and this journal article.

Results

Sample Characteristics

There were 288 participants, who met the criteria and had children diagnosed with either autism (38.5%, n= 111); ADHD (32.6%, n = 94); and autism+ADHD (28.8%, n= 83). The children in the sample were predominantly male (74.7%, n =215) followed by female (22.6%, n = 65) and nonbinary (2.1%, n = 6) with a mean age of 9.5 years. On average the autistic children were diagnosed around 5.25 years (7.3 years for the autism diagnosis for the autistic + ADHD children) and 7.4 years for ADHD children (6.8 years for the ADHD diagnosis for the autistic + ADHD children).

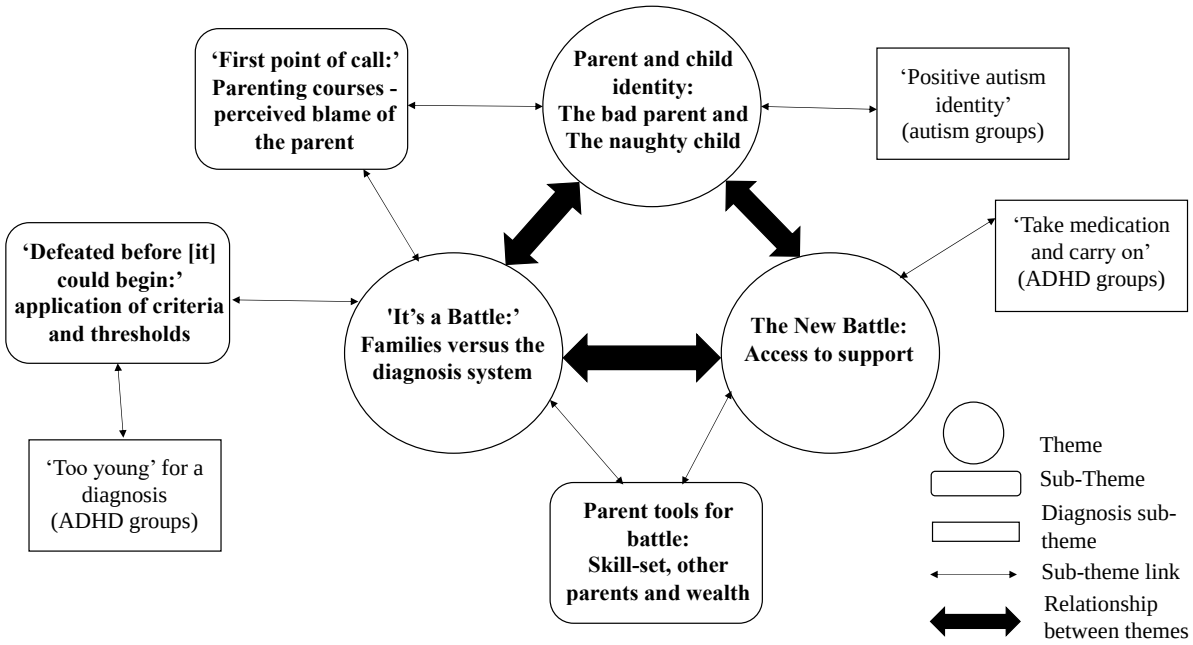
Mothers (94.8%, n = 273) were more likely to have completed the survey than fathers (2.4%, n = 7) or legal guardians (2.8%, n = 8). The parents who completed the survey were more commonly NZ European (69.4%, n = 200) compared to Māori (22.2%, n = 64) or Pacific People and other cultural backgrounds (8%, n = 23), had completed university (42.4%, n = 131) and were in a household income bracket between $NZ 50,000 - 99,000 (36.5%, n = 105).

Supplementary figures 1-3 (Appendix F) show the rate of response and approximate length of responses to the three questions. There were a total of 694 responses across the three questions totalling 19,509 words, which ranged from one word to 500+ word responses. There were 26 participants who did not answer any of the three questions (n = 11 ADHD; n = 7 Autism; n = 8 autism + ADHD participants). There was no significant difference in the rate of 'no response' or length distribution across the three diagnostic groups. The responses produced a rich and varied qualitative data set.

Thematic Analysis

Analysis of the data led to the construction of three major themes: 1) it's a battle, 2) parent and child identity, and 3) the new battle. The relationship between these three themes and subthemes are illustrated in Figure 5.1. The answer to the question "what was helpful?" was inverted by 25% of respondents, who answered this negatively, such as '*Nothing, we really didn't get any help*' (ADHD).

Figure 5.1. Thematic map of parents' experience of the diagnostic process for their children.



Theme 1: 'It's A Battle': Families versus the diagnosis system

This first theme, which describes the battle and struggle parents of children with Autism and/or ADHD had with the diagnosis system, is made up of four subthemes, two of which also apply to another theme. These are: (i) *defeated before it could begin*, which pertains to parents' perception of diagnostic-delay due to restrictive criteria; (ii) *too young for a diagnosis,* relating to a particular restrictive age criteria, which applied to obtaining an ADHD diagnosis (iii) *first point of call,* (described in Theme 3: Parent and child identity) and (iv) *parent tools for battle* (described in Theme 2: The new battle).

Parents described battling or fighting within a system for a diagnosis for their child, as being in a '*long battle*' (Autism + ADHD) and the diagnosis system as '*broken*' (Autism), *'a nightmare'* (ADHD) and '*like walking to hell and back.'* (Autism). Parents described an 'us' versus 'them' situation where they had to constantly '*fight to get him seen*,' (Autism + ADHD) or to better co-ordinate within the system; '*I fight every single day to stay on top of the people/agencies [...] to not get lost in the system.'* (Autism + ADHD). When parents expressed that they had a positive experience they were compelled to acknowledge this as luck, or an exception to the normal experience of having to fight for their child*: 'For other families, I hear how much harder it is, so I consider our experience more of an outlying data point than indicative of the normal diagnostic process in New Zealand'* (Autism).

'Defeated before [it] could begin': Application of Criteria and Thresholds

The application of criteria for assessment and diagnostic thresholds were a key component of the parents' battle for access. Parents were told their child was 'too mild,' 'too social' or 'too young' to get a diagnosis and were disappointed by not being given alternative options after their child failed to meet criteria. The resulting diagnosis-delay left parents feeling demoralised and shut out: '*The public system said he was not bad enough therefore would not help us'* (Autism + ADHD). This created a fait accompli and some families felt beaten when advised not to try for assessment in the public system; '*GP thought the wait in the public system would be too long and threshold of impairment too high to be accepted for assessment'* (ADHD). Meeting the threshold of 'mild' became something negative that often led to denied access, rather than acknowledgement and justification for accommodation for the needs of the child and family.

Sometimes professionals decided to delay a referral or diagnosis due to thresholds and criteria: '*while you want to be thorough, delaying a diagnosis does not help children and families'* (Autism). In some cases professionals applied their own criteria, and did not

diagnose because they believed *'labels are bad'* (Autism), or because they had a lack of *'belief'* that there was an issue (ADHD). In other cases, parents described being stonewalled by clinicians who declared that their child did not fit the diagnostic criteria, but who did not offer counter evidence or only spurious evidence. For example, a clinician was reluctant to diagnose because a child made eye-contact so was 'not autistic' (Autism + ADHD). Narrow criteria and subjectively applied thresholds denied or delayed diagnosis and lengthened the 'battle.' This experience was in contrast to helpful professionals, who validated concerns, explained the diagnosis in relation to the child, and offered tangible help, such as, describing the doctor *'who explained that' our child's brain just worked differently and explained it in a way for us all to understand'* (ADHD)

'Too young' for a diagnosis (ADHD groups)

Parents in the ADHD groups encountered the professional's barricade of being told their child was 'too young' for a diagnosis: '*We kept getting told he's too young... age is sometimes irrelevant when there is obvious signs'* (Autism + ADHD). Parents were upset because this decision delayed diagnosis, reduced time for early support and left the family in limbo: '*It is very, very hard to get a diagnosis for young children and all the research shows the earlier you get this the better'* (ADHD). Parents described how they were thwarted when told to wait until the child turned six or had started school and that this led to a feeling *'of being defeated and overwhelmed'* (Autism + ADHD). Delays also meant parents were not able to access support and had to start the process again: '*because he was too young I couldn't get any support - We were discharged from service then had to get another referral when he turned 6. Then the wait list and re-referral process took another year to get through'* (ADHD).

Theme 2: The New Battle: Access to Support

The second key theme was that, following their initial battle (getting a diagnosis), parents reported having to start a new fight to get support for their child. This theme has two subthemes: (i) *Parent tools for battle* (also relates to Theme 1); and (ii) *Given medication and told to carry on* which described the quandary parents felt around medication being given in isolation to children diagnosed with ADHD.

This second and new battle was described by some parents as harder than their initial fight for diagnosis: '*The diagnosis part was easy. It's getting the required support afterwards that is hard'* (Autism). The support was described as concealed and costly; *'If she needs any support now, we have to find it and fund it ourselves, which we can't really afford to do'* (ADHD). Parents described being disappointed and let down because the purpose of diagnosis was to get support for their children, so when no support was forthcoming the diagnosis seemed '*pointless*' (Autism), for '*everyone else*' (Autism) or that diagnosis was impractically treated as a '*magic wand*' with no other support (Autism + ADHD).

Accessing support in the education system was challenging and parents encountered many obstacles including a lack of visibility, insufficient learning support, and limited teacher capacity. Some parents drew attention to the criteria for school support, including accessing teacher aides in the classroom, explaining that unless the child needed '*help with toileting*' (Autism), or the child was '*extreme and throwing chairs at teachers*' (Autism + ADHD) then there was no support from the education sector. One parent explained that the teacher aide hours had recently been cut to serve more children but with shorter amounts of time (ADHD).

Parents also identified that access to education support was adversarial in nature and seemed ad hoc, uncoordinated, and 'un-planned'(ADHD). They noted that support dropped off at the start of school enrolment rather than increased (Autism) and that there was a gap

between diagnosis and support at school (Autism). One parent explained how they were caught between organisations over the responsibility for support; '*Hospital thinks schools should support and schools think its hospital's role to support. Parents are stuck in the middle'* (ADHD). The limited support was also not parent and family-focused and parents lamented a lack of parental counselling '*to come to terms with diagnosis,'* (Autism) and that overall *'the system is not at all prepared to support us.'* (Autism + ADHD).

Parent tools for battle: Skill set, other parents, and wealth

Whether parents were starting their first battle with the system for diagnosis or beginning a new (second) battle with the system for support their experiences were shaped by the assets and tools they had at their disposal. Parents who were able to draw on previous knowledge, and knew how to research their child's condition and navigate health systems were more prepared for their fight: '*I feel like we only managed to get through the process because we are well educated and persistent.'* (ADHD). The parent skill-set was seen as essential due to the level of difficulty required to navigate the pathway to diagnosis: *'It was very difficult navigating the diagnosis pathway. My background is in the heath sector and I consider myself to be reasonably well informed.'*(ADHD). Parents were acutely aware and concerned for other parents who might not have the necessary skill-set: '*Following the diagnosis path to get help without having these tools and knowledge would be even harder'* (Autism).

Parents' sense of empowerment that came with having assets was sometimes undermined during professional encounters. One parent explained that after a psychology degree and multiple parent courses one specialist told her *'"that sounds like parent diagnosis," actually, I know my son best'* (Autism + ADHD). Parents sometimes felt better informed than professionals, with particular skill-sets such as in understanding girls with

ADHD or autism: '*There is not enough knowledge in the medical profession about autism in girls and how it presents differently. It's draining.'* (Autism + ADHD).

Another important tool parents used in their fight for diagnosis was support and comradery from other parents who became allies and shared information about how to navigate the system: '[We] *only knew what to do (e.g. ask GP for a referral to specialist) as my friend had recently been via the process for her son.'* (Autism + ADHD). Parents described how information, support, and helpful agencies were '*very much word of mouth from other parents,'* (Autism + ADHD), which was *'invaluable'* (Autism). This skill-set sits outside of the system with a clear delineation between 'us' (parents of children with diagnoses) and 'them' (the systems and professionals involved along the diagnosis pathway).

The other tool for fighting the system was the personal financial resources parents used to access professionals and educational support in the private system. Parents often acknowledged how lucky they were to be able to go private: '*Expensive but we are lucky that's an option for us.'* (Autism). Parents lamented the cost, but explained the private system fulfilled their expectations: *'If you don't come from a family with means to pay for assessments, tuition and support required you are left behind by our system.'* (Autism + ADHD)

'Given meds and carry on' (ADHD and autism + ADHD)

A key subtheme in the 'new battle' was that support for children with an ADHD diagnosis often consisted solely of being given medication: '*very little support given for practical strategies [...]. Basically given meds. and carry on*' (ADHD). One parent of a child with both autism and ADHD contrasted the support after diagnoses by commenting that whilst no support, other medication had been offered for ADHD there and been*,' information overload for autism'* (Autism + ADHD). Parents of children with ADHD also shared how

medication had been '*life changing*' (ADHD) with their child being able to engage in the classroom.

Theme 3: Parent and Child Identity: The Bad Parent and the Naughty Child

The third key theme in the parents' experience of diagnosis and support concerned parent and child identity. Parents' competence and children's behaviour were often blamed prior to diagnosis and a label led to a shift in blame and identity. This theme is connected to both 'Battle' themes and is made up of two subthemes: (i) *'First point of call'* which pertains to the way parenting courses are offered (also relates to Theme 1) and (ii) 'Positive autism identity' which relates to an identity specific to the autism diagnosis.

Prior to diagnosis parents carried responsibility and blame for their child's behaviour, making them particularly vulnerable to directly or indirectly implied parenting criticisms. Parents described being blamed by other parents and professionals and '*Being made to feel like a horrible parent'* (Autism + ADHD). They felt their ability and expertise questioned and critiqued by other parents and services: '*It is a very, very hard road with judgement from other parents and most of all the agencies who think it's your parenting*' (Autism + ADHD). Professional sometimes appeared to locate the problem in the child or parent rather than in the condition and accompanied this with dubious parenting advice such as a doctor telling a parent it was *'a testosterone surge and to go home and be a parent'* (ADHD), or that *'maybe he just needed a smack and more discipline'* (Autism). The parents were also denied a referral to a specialist, and this created a diagnosis-delay.

For some parents, receiving a diagnosis for their child led to validation and a change in identity for parents. They experienced relief due to a reduction in social stigma and described how others no longer judged them as bad parents: '*It no longer felt like a helpless situation due to bad parenting - people believed me finally'* (ADHD). Equally, upon diagnosis there was a change in identity and a shift in blame away from their child which was

a relief for parents; *'Realising we weren't bad parents, our child wasn't naughty.'* (Autism). The diagnosis enabled parents to actively reframe their child's identity at school and home, and rationalise their child's behaviour.

'First point of call': Parenting courses - perceived blame of parent

Parents were often offered parenting courses by professionals. This was perceived by parents as confirmation of their, sometimes self-imposed, 'bad parent' identity: '*They blame you initially. Send you to parenting courses. Many different parenting courses. Then eventually after a few years on a wait list you see a specialist.'* (Autism + ADHD). This subtheme was also shared with the first theme 'It's a battle' by the timing of these parenting courses, which appeared to act as gatekeepers for accessing a diagnosis system and were perceived as an affront to parents: '[organisation]*'s lack of interest in a person unless they do a parenting course first is pathetic.'* (ADHD)

Although some parents could see the benefit of these courses, they nevertheless wanted the timing to be reconsidered: '*I understand there needs to be a focus on parenting in some respect, but that shouldn't be the first point of call.'* (Autism + ADHD). Parents were often referred to parent education programmes instead of being referred for diagnosis, because professionals perceived this as a form of helpful support for parents. Parents highlighted the contradictory message inherent in the offering of parenting courses as support; '*Although there is a strong push toward parents not having done anything wrong etc., but then the first thing offered is a parenting course... This is an issue'* (ADHD). By choosing parenting courses as a first course of action it was perceived that professionals ignored the competence and skill-sets of parents and undermined parents self-efficacy; '*It was also humiliating being forced to do a parenting course as I have a background as a parenting educator!'* (Autism + ADHD). The courses were also criticised for not specifically

addressing parenting of neuro-diverse children: *'makes people feel bad and many of the strategies don't work for kids with ASD / ADHD.'* (Autism + ADHD).

Positives of an Autism Diagnosis (Autism and Autism + ADHD)

A second subtheme, related to parent and child identity, was that of a positive autism identity. Some experienced the opportunity to reposition their identity as parents of an '*authentically autistic*' child (Autism), advocating the involvement of autistic adults in information and organisations: *'Nothing about us, without us'* (Autism).

This second identity shift came from a number of different sources, including wider experience with autism in the family, autism community groups and sometimes professionals. The community groups saw labels as empowering and parents lamented how autism had been '*medicalised and problematised*' (Autism) during the diagnostic process. Some professionals were described as having rigid concepts of normality and typicality, others encouraged parents to see autism through an inclusive lens and with positivity; '*[Professional] treats autism as a normal variation and doesn't stigmatise it.*' (Autism + ADHD).

Discussion

The reported experiences of obtaining a childhood diagnosis for autism and ADHD were similar with common themes expressed by parents across diagnoses. Many parents experienced the diagnostic process as a battle, which was primarily motivated by the impetus to obtain support for their child. The analysis of the parents' experiences shows that delays in diagnosis might be the result of factors including opaque systems, wait-and-see models, and requirements for parenting courses before diagnosis. The diagnosis appeared to have a particular benefit in allowing parents to reframe their parental ability and the behaviour of the diagnosed child to a more positive and less guilt-laden identity.

The findings from our study show that the 'battle' analogy was omnipresent throughout the process and informed each stage. Across all parents in this analysis, there was

disappointment that they had to start a second fight for support after diagnosis, particularly within an education setting. These findings align with those of Daniels et al. (2021), who studied clinicians' understanding of parents' experience of the journey to the autism diagnosis in United Kingdom, and created a subtheme ‘parental battle for services’ where clinicians used the battle analogy to describe their perspective of parents' ‘fight’ for support.

Parents' perception of parenting courses as part of this ‘battle’ was perhaps surprising because parenting courses are recognised as an evidenced-based support (Prata et al., 2018). As a ‘first point of call’ before diagnosis, these courses might rule out more mild issues and thus, reduce wait times, patient load, while also potentially benefitting the child and parent (Risley et al., 2020). There a number of recognised benefits of parenting courses, such as a reduction in school and relationship issues, and a sense of greater parent self-efficacy and well-being of the family (Leijten et al., 2018; Modesto-Lowe et al., 2008; Prata et al., 2018). However, parents in the present study viewed being offered parenting courses as evidence that their parenting was being blamed or implicated. The theme of parent and child identity indicates that parents may well be primed for this thinking both from internal and social pressures at the outset of the process to diagnosis. Whilst Kasilingam et al. (2021) describe how 22% of New Zealand parents of autistic children specify parenting courses as the extra support that they would like, the question was posed after diagnosis. Parents may have experienced the referral differently had they been offered the courses as a support after diagnosis and specific to the child's diagnosis.

The comparison of the three diagnoses in the present study highlighted unique themes for the ADHD group with the use of medication for treatment and with the application of the ‘too young’ for diagnosis criteria. In contrast to the ADHD groups, the autism only group did not report being told their child was too young for a diagnosis, perhaps because the message about early diagnosis being possible from age two is becoming more well-known

(Waddington et al., 2021). Halperin and Marks (2019) identify a number of reasons why practitioners are reluctant to diagnose ADHD in preschool including that the symptoms of ADHD are commiserate with typically developing preschool children, multi-informant assessments are more difficult to establish and that early childhood symptoms are not well established. Although there is growing body of research on the diagnosis of ADHD in pre-schoolers (Wallisch et al., 2020; Wigal et al., 2020), in the current clinical context the explanation of ‘too young’ is understandable, but nonetheless provides a mismatch between parents’ options for early diagnosis, intervention, and support.

A unique theme for the autism groups was that parents called for an introduction to thinking about a positive autism identity to occur along the diagnostic pathway. There is an important trend in the research literature and in community groups, away from ‘having autism,' and autism being framed using a bio-medical deficit-focused model, to ‘being autistic’ and the acknowledgement of a more social or neurodiversity affirming identity (Anderson-Chavarria, 2021). A more positive framing of ADHD abilities in the research literature is now occurring (Moore et al., 2021; Sedgwick et al., 2019), but there seems to be less advocacy and sense of a positive ADHD identity compared to autism within the community of parents' surveyed in the current research.

The current research was limited in a number of ways. The survey represented a cross-section of New Zealand population based on the average highest education and income of the sample (OECD, 2019); however, certain groups may have been under-represented in responding to the survey. The majority of participants were mothers, which limited the voices of fathers, and a more gender-balanced sample might be considered for future research. This survey did not analyse parents' responses to the diagnosis process over time or contextualise particular disparities, such as rural, cultural and low socio-economic groups. A further consideration is that parents of children in the survey answered if there child was between

one year to eighteen years of age, which means that retrospective remembering of the process might differ between the different points of diagnosis and in relation to the current age of the child. In addition, the opt-in nature of the survey may have meant that participants felt compelled to answer if they were particularly aggrieved or if they had a negative diagnostic experience. This creates a less favourable picture of the process. An open-survey is also restrictive because it does not allow researchers to follow-up or probe further in understanding the diagnostic experience.

Support organisations and professionals might wish to consider working more collaboratively with parents, to acknowledge parental guilt and to signal upfront the process from diagnosis to support options. Understanding the centrality of parent and child identity might encourage professionals to be more sensitive around their interactions. An empowering of autism and ADHD identity might help parents along the diagnostic pathway. The criteria set of being 'too young' for ADHD families should be examined and the timing and appropriateness of recommending parenting courses reviewed. This might involve addressing parent experiences of the courses acting as a gatekeeper to diagnosis, the applicability of the courses to neuro-diverse children and the implied assumptions about parenting competence and expertise. The similarities of experience across the three diagnostic groups suggests that a wider parent community group might draw on greater expertise about access, support and advocacy of neurodiversity. Future research might examine further similarities across childhood neurodevelopmental conditions and those with co-occurring diagnosis experience.

Conclusions

This study has shown that, during the diagnostic pathway, parents of children, who have either ADHD, autism or autism and ADHD, experienced a similar battle to get a diagnosis for their child and access support for their child and family. Many of the issues found in this research, were similar to other research examining the experience of autism

diagnosis, yet were found also to be applicable for ADHD and a co-occurring diagnosis. However, there were specific themes for the particular diagnoses, such as parent's desire for the empowering of a positive autism identity, and the application of a 'too young' criterion for parents with ADHD children. Parents were sensitive to perceived parental and child blame in interactions along the diagnosis process, which meant parenting courses were problematic when offered prior to diagnosis.

CHAPTER 6

STUDY 3: An exploration of clinicians' approach to complex neurodevelopmental cases

Chapter Note

This chapter has been revised and resubmitted to the *Journal of Paediatrics and Child Health.* Dr Hannah Waddington helped with the design, analysis and editing. Prof. Andrew Whitehouse and Dr. Kelly Carrasco assisted with design and editing. I designed, conducted and wrote this article and it is entirely my work aside from the input specified above. Aside from changes to ensure consistency of formatting with the rest of the thesis, this chapter is identical to the article that has been submitted for consideration.

Abstract

Aim: This study examined clinicians' initial approach to hypothetical complex neurodevelopmental cases. **Methods**: Clinicians listed possible assessments, services and diagnoses they might consider in response to vignettes describing a 4-year-old and 8-year-old child. The wide range of behavioural concerns present in the vignettes were suggestive of a number of different neurodevelopmental co-occurring symptoms. **Results**: Twenty-nine clinicians' responses indicated that case complexity does not follow a predictable and universal clinical pathway, but instead multiple possibilities were considered. The number of possibilities indicated that considerable time and resources may be required to diagnose complex neurodevelopmental cases and that some aspects of the referral were of particular concern. The clinicians suggested a number of support services, which were most commonly referrals to other organisations. **Conclusions**: Clinicians have multiple considerations when considering a referral, which include possible assessments, immediate and long-term support and referral concerns which might indicate safety or current well-being issues, that might take precedent over clinical diagnostic concerns.

Keywords: Diagnosis, complex cases, vignette, neurodevelopmental conditions, co-occurrence, autism, ADHD

Brief points:

- Previous research examining the process of diagnosing neurodevelopmental disabilities has generally specified the intended diagnosis (e.g. autism) at the outset, by contrast, this study sought to examine the challenges for clinicians when presented with a hypothetical vignette case for a 4- and 8-year old with symptoms indicative of multiple neurodevelopmental conditions.
- Clinicians considered multiple assessments, diagnoses and possible services in relation to both vignettes.
- Recommendations are made for reconciling the discrepancy between the pressure on clinicians to quickly and accurately diagnose neurodevelopmental conditions and the lengthy assessments and multiple considerations of co-occurrence.

Neurodevelopmental conditions are characterised by a range of behaviours that are due to atypical brain development (American Psychiatric Association, 2013). Diagnosing neurodevelopmental conditions, as defined by the categorical Diagnostic and Statistical Manual 5th edition (DSM 5), can often be challenging due to symptom variability across individuals, developmental changes over time, the presence of "borderline" cases and the frequency of co-occurrence (Astle et al., 2021). The challenges and considerations of co-occurrence are also compounded by pressure on clinicians to reduce waiting times and achieve accurate diagnosis so that families can access early support (Whitehouse et al., 2021). Two particular neurodevelopmental conditions, autism and attention-deficit/hyperactivity disorder (ADHD), demonstrate these challenges. Autism is often described as difficult to diagnose, even in the absence of ADHD and other symptoms, due to behavioural and

biological heterogeneity and changes over a developmental period (Taylor et al., 2021). The same is true of ADHD diagnosis (Halperin & Marks, 2019). The prevalence estimates for co-occurring autism and ADHD are between 30-70 % (Joshi et al., 2017).

The difficulties in diagnosing co-occurring autism and ADHD are reflected in a delayed autism diagnosis of over a year later on average when ADHD is present (Sainsbury et al., 2022). This delay may be partially due to "diagnostic overshadowing", where autism symptoms are masked by a child's more apparent ADHD symptoms (Gipson et al., 2015; Soke et al., 2018) or "diagnostic substitution", where a child is given a diagnosis as indicated by symptoms presentation, but a co-occurring diagnosis or other developmental disability covers a more comprehensive symptom profile of the child (Heyman et al., 2022). Clinicians may also find one diagnosis that fits and fail to consider further diagnoses (Gipson et al., 2015; Soke et al., 2018). Case-complexity is usually defined by the presence of co-occurring symptoms, which complicate a differential diagnosis or diagnoses (Heyman et al., 2022).

The presence of other conditions and symptoms such as anxiety, behavioural issues, and sleeping difficulties across these two conditions can further complicate the presentation (Soke et al., 2018). Autism and ADHD both have high rates of co-occurrence with other conditions. Specifically, 95% of autistic children are estimated to have other symptoms or diagnosed conditions (Soke et al., 2018) and over 52% of ADHD children are also diagnosed with a co-occurring condition (Jensen & Steinhausen, 2015). Indeed, 8-year-olds diagnosed with autism in the USA had an mean of 4.9 co-occurring conditions or symptoms (Soke et al., 2018). These co-occurring conditions and symptoms including hyperactivity, elopement/wandering, sleep difficulties, ‘temper tantrums’, and mood changes often cause the most stress and concern for parents of autistic children, and these parents may therefore be more likely to report these behaviours to a professional than the core characteristics of autism (Anderson et al., 2012; McLaughlin et al., 2020; Pereira-Smith et al., 2019).

Diagnosis of co-occurring neurodevelopmental conditions is complex and may be challenging for clinicians. Research into professionals' perspective on autism diagnosis often begins from the a priori assumption that the individuals being assessed are autistic or suspected to be autistic (Taylor et al., 2021). However, this study sought to examine the difficulty for clinicians when presented with a complex case with multiple neurodevelopmental symptoms, including ADHD and autism symptoms. This was presented as vignette referrals for a 4- and 8-year-old child.

Methods

Ethical Clearance and Informed Consent

Ethical approval for this study was provided by the Human Ethics Committee at Victoria University of Wellington, New Zealand [Approval number 28993]. Participation in the study was both voluntary and anonymous. Participants were asked not to include any information which would make them identifiable. Participants could also choose not to answer questions using a "prefer not to say" option.

Participants

Participants were eligible for inclusion if they were involved in the diagnosis of neurodevelopmental conditions in children in New Zealand (hereafter, clinicians). Convenience sampling was used, as participants were recruited by an email sent by the New Zealand Psychologists Board, district health boards and paediatrics groups in New Zealand.

Materials

The vignettes used in this study were based on research into behaviour that is likely to co-occur across neurodevelopmental conditions (Heyman et al., 2022; Soke et al., 2018; Yorke et al., 2018) as well as an amalgamation of cases experienced in clinical practice by the authors. The symptoms in the vignette were designed to illustrate a complex case

presentation rather than clear examples of DSM 5 criteria and, as such, there was no one "correct" response (American Psychiatric Association, 2013) (see tables 6.1 and 6.2).

The survey was hosted on Qualtrics from the 15th of March 2021 until the 20th October, 2021. The survey was revised after consultation with two paediatricians and a psychologist. The questionnaire had 17 questions separated into 3 sections related to: (i) demographic characteristics, (ii) the vignette referral for a 4-year-old, and (iii) the vignette referral for an 8-year-old.

An initial screening question asked whether the clinician diagnosed children in New Zealand. If the participant answered "no," they were ineligible to continue. The demographic characteristics section included questions about ethnicity, profession, length of time practicing, primary location of training, sector of work (public/private), and specialty interest.

Then, clinicians were presented with the 4-year-old vignette, followed by the 8-year-old vignette (see Tables 6.1 and 6.2). Clinicians were then asked to rate the extent of their

Table 6.1.

Vignette of four-year-old child with behaviour possible alignment to DSM 5 criteria.

Vignette referral text	Behaviour short hand description	Possible alignment with DSM criteria for Autism	Alignment with DSM criteria for ADHD	Suggestive of other neuro-developmental conditions	Categorisation
Delayed speech; short course of speech and language therapy (SLT). He is now speaking in three word phrases	Language delay	No	No	Language disorder	Other
He can name all his colours, count to 5, but jumbles his numbers after 5.	Cognition	No	No	Global developmental delay	Other
Parents separated when he was 3 years old.	Parent separation	No	No	No	Other
He does not participate well with the rest of the children, particularly during mat time	Lack of Participation	A3. Deficits in developing, maintaining, and understanding relationships	1c. Often does not seem to listen when spoken to directly 2b. Often leaves seat in situations when remaining seated is expected	Anxiety disorder	Autism and ADHD

He will latch on to one adult or child for the day and direct them by lifting their hand to what he wants	Using other's as a tool and latch on to a person (Person as tool)	A1. Deficits in social-emotional reciprocity	No	No	Autism
He likes the dinosaurs and he will place them in different parts of the centre	Placing objects	B1. Stereotyped or repetitive use of objects.	No	Obsessive compulsive disorder	Autism
and he sometimes becomes very upset if the dinosaurs are then moved, which can lead to him throwing things.	Becomes very upset (upset)	B2. Insistence on sameness, inflexible adherence to routines	No	Obsessive compulsive disorder	Autism
He will often try to escape the childhood centre, seeing it as a game by taking any moment to run through the gate and run off down the driveway	Elopement		2c. Often runs about or climbs in situations where it is inappropriate		ADHD
He likes roaring loudly like a dinosaur	Loud vocalisations	No	2D. Often unable to play or engage in leisure activities quietly		ADHD
He likes hiding under tables	Hiding	No	No	Anxiety Disorders	Other

*Note: Table is not an exhaustive list and behaviour only considered possibly suggestive of the criterion.

Table 6.2.

Vignette of eight-year-old child with behaviour possible alignment to DSM 5 criteria.

Vignette referral text	Behaviour short hand description	Alignment with DSM criteria for Autism	Alignment with DSM criteria for ADHD	Suggestive of other neurodevelopmental disorders	Categorisation
Difficulty sleeping at night, and his mother reports poor sleep from an early age	Difficulty sleeping	No	No	Sleep disorder	Other
Cognitive assessment which raised no concerns with a reported IQ score in the average range	Average IQ	No	No	No	Other
He has trouble making friends at school, and he has started a social skills program with his teacher and teacher aide in a small group	Difficulty making friends	A3 Deficits in developing, maintaining, and understand relationships	No	No	Autism
The teacher reports that he often forgets what task he should be working on	Forgets what to do	No	1b. Often has difficulty sustaining attention in tasks or play activities 1d. Often does not follow through on instructions and fails to finish schoolwork		ADHD

Instead often shouts a swear word to be funny for his classmates	Swearing	A1. Deficits in social-emotional reciprocity	2i. Often interrupts or intrudes on others.		Autism and ADHD
He has trouble following classroom instructions	Difficulty following instructions	No	1d. Often does not follow through on instructions		ADHD
He does not appear interested in much at school, but at home his mother reports that he loves playing Lego	Low interest	A1. Deficits in social-emotional reciprocity.	1f. Often avoids, dislikes, or is reluctant to engage in tasks that require sustained mental effort		Autism and ADHD
He gets told off frequently for his voice which is either shouting or a whisper.	Shouting/whispering.	A2. Deficits in nonverbal communicative behaviours used for social interaction	2f. Often talks excessively	Tourette Syndrome	Autism and ADHD
The other children typically avoid Thomas	Not socially accepted.	No	No	No	Other
He tends to move from his seat to get closer to his classmates to see what they are doing	Leaves seat.	No	2b. Often leaves seat in situations when remaining seated is expected 2i. Often interrupts or intrudes on others	No	ADHD

*Note: Table is not an exhaustive list and behaviour only considered possibly suggestive of the criterion.

concerns related to ten identified aspects of the vignette (e.g. language delay) across a five-point Likert scale from "not at all important" to "important." The following open-ended questions were asked: i) what assessment(s) would you consider for this child; ii) what age might you recommend the assessment(s) be done; iii) what diagnoses might you be considering for this child; and iv) would you consider recommending any services.

Data Coding and Analysis

The first author matched referral concerns against the DSM-5 criteria (American Psychiatric Association, 2013). This was checked by the second and last author and disagreements were resolved. Descriptive statistics were used to illustrate the sample demographic characteristics, the median level of concern across clinicians for each referral behaviour, and the percentage of clinicians who suggested different possible assessments, diagnoses, and support services. Specific named assessments (e.g., Behaviour Assessment for Children - BASC III) were coded into categories (e.g., general developmental assessments).

Results

Sample Characteristics

Thirty-two clinicians participated in the survey. Of these, 3 clinicians only completed the demographic questions, and were excluded. Table 6.3 provides the demographic characteristics for the remaining 29 participants. The clinicians were predominantly New Zealand European, trained in New Zealand, and had over 10 years of experience.

Table 6.3.

Demographic characteristics of participants (n = 29).

Demographic characteristic	Total n (%)
Ethnicity	
NZ European	21 (72.4)
Māori	3 (10.3)
Asian	3 (10.3)
Prefer not to say	2 (6.9)
Profession	
Paediatrician	15 (51.7)
Psychologist	11 (37.9)
Other (psychiatrist, GP, paediatric nurse practitioner)	3 (10.3)
Experience	
10 years +	24 (82.8)
Under 10 years	6 (20.7)
Location of training	
New Zealand	27 (93.1)
Other	2 (6.9)
Sector	
Private	6 (20.7)
Public	17 (58.6)
Both (private and public)	6 (20.7)
Specialty	
No or prefer not to say	10 (34.5)
Neurodevelopmental conditions, ADHD or autism	6 (20.7)
Other	13 (44.8)

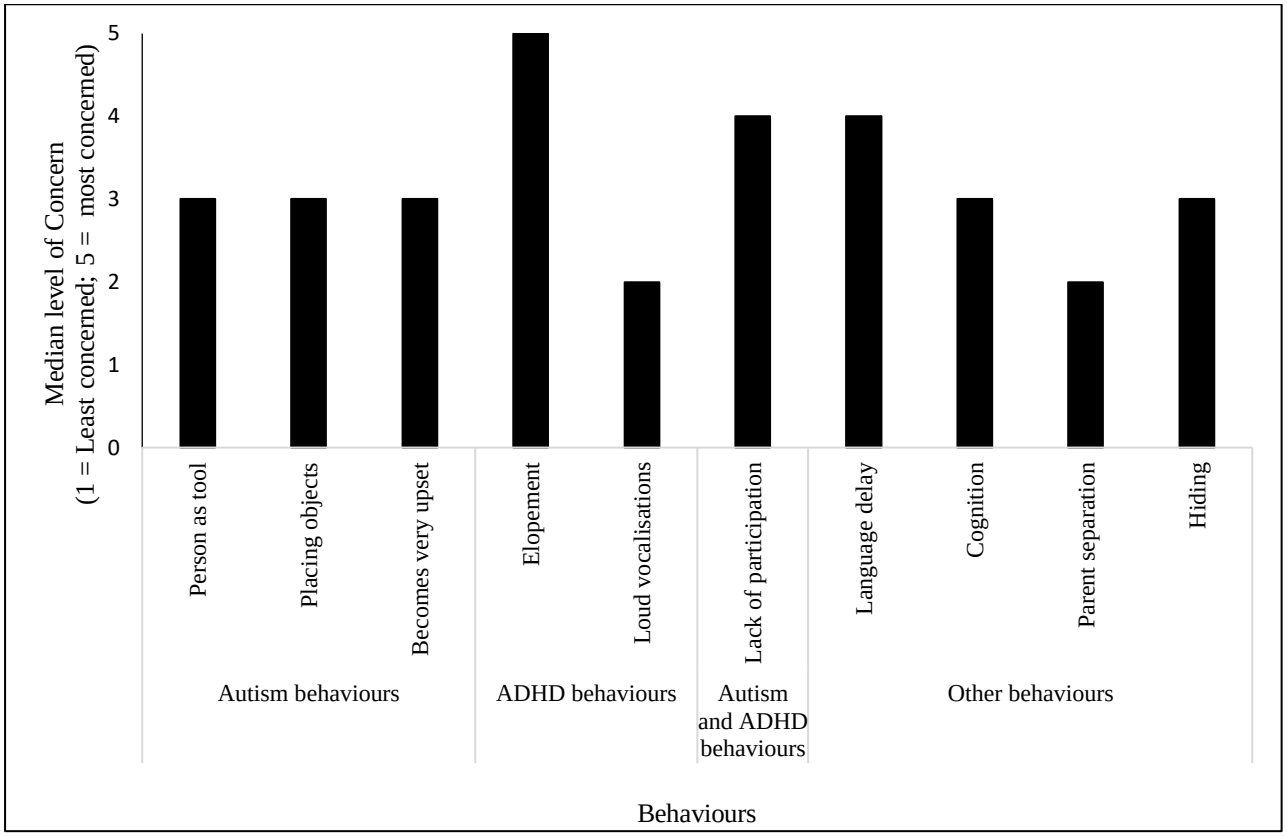
Clinicians' Responses to the Vignette Referral of a Four-year-old

Referral Concerns

Figure 6.1. shows a bar graph of clinicians' median concerns across the various behaviours presented in the vignette. The behaviours of greatest concern across clinicians were elopement, followed by language delay and lack of participation.

Figure 6.1.

Clinician's median areas of concern regarding a vignette of a complex case (4-year-old)



Timing and Nature of Assessment

Table 6.4 indicates whether the clinicians would recommend immediate or delayed assessment in relation to the vignette. Three participants indicated they would not assess straight away. The other 26 opted for an immediate assessment. Table 6.4 also shows the different assessments that the clinician would consider. The clinician listed ten different assessment possibilities with the two most common being an observation and a developmental assessment. The average number of assessments listed by each clinician was

three. Clinicians frequently suggested that interviews were conducted alongside either an observation or a developmental assessment. Multiple practitioners also indicated that more assessments would follow initial findings.

Table 6.4.

Clinicians' suggestions regarding possible assessments, diagnoses and interventions of complex case described in a vignette of a four-year-old (n = 29).

	Total n
Possible Assessment timing	29
Immediately (age 4)	26
Later (6 years +)	3
Possible Assessments	
Developmental assessment	14
Autism assessment (ADOS etc.)	13
Observation (at home, at ECE)	12
Interviews (Parent, ECE teacher)	11
Cognitive assessment	9
Physical and medical assessment	5
Speech and language assessment	5
Social communication assessment	4
ADHD (Conners etc.)	3
Assessment referral to colleague	3
Trauma history	3
Other assessments guided by findings	5
Possible Diagnoses	
Autism	19
Global delay	10
Attachment condition/difficulties	10
Language delay	7
Trauma	7
Intellectual difficulty	6
ADHD	3
Medical/genetic diagnosis	3
Specific learning disability	2

Anxiety	2
Foetal alcohol spectrum condition (FASD)	1
Too early to speculate on diagnosis	6
Other unspecified diagnoses	5
Possible Services/Interventions	
Speech and language therapy	10
Early intervention teacher	9
Support for early childhood education setting	6
Play therapy	6
Educational psychologist	5
Refer to multidisplinary team (child development service)	4
Social worker	2
Audiology support	2
Parenting course	1

Possible Diagnoses

Table 6.4 indicates the diagnoses that clinician considered for the 4-year-old. Eleven different possible diagnoses were suggested, the most common of which were autism followed by global delay and attachment difficulties.

Recommended Services

Table 6.4 indicates the possible services that clinicians would recommend. Fourteen different services were recommended. The average number of services recommended by each clinician was two. The two most common were to continue with speech and language therapy and to gain support from an early intervention teacher. All of the services involved a referral to another programme or colleague specialty.

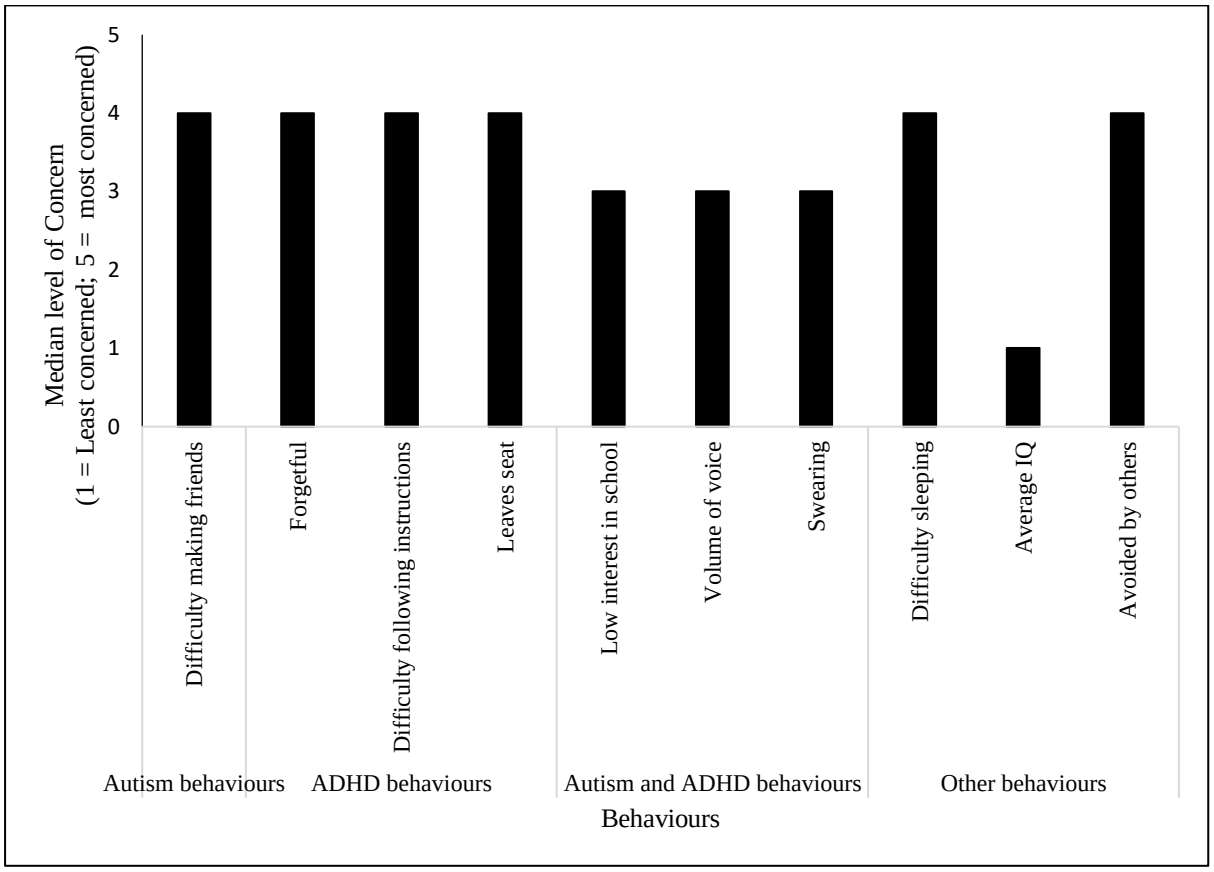
Clinicians' Responses to the Vignette Referral of an Eight-year-old

Referral Concerns

Figure 2 shows a bar graph of median concerns indicated by clinicians across the various behaviours presented in the vignette. The graph shows that multiple factors were a concern leading to a number of different symptoms to address as a clinician.

Figure 6.2.

Clinicians' median areas of concern from a vignette of a complex case (8-year-old)



Timing and Nature of Assessment

Table 6.5 indicates that all clinicians opted for an immediate assessment, except for one who stated it was not applicable. Thirteen different assessment possibilities were listed. The average number of assessments listed by each clinician was three. The two most common assessments were an interview and an ADHD assessment. Clinicians frequently

suggested that interviews were conducted alongside an ADHD assessment. Multiple practitioners also indicated that more assessments would follow initial findings.

Table 6.5.

Clinicians' suggestions regarding possible assessments, diagnoses and interventions of complex case described in a vignette of an eight-year-old (n = 29).

	Total n
Possible Assessment Timing	
Immediately (age 8)	28
Assessment not applicable	1
Possible Assessments	
ADHD (Conners etc.)	21
Interviews (Parent, teacher)	13
Autism assessment (ADOS etc.)	8
Observation (at home, at school)	8
Developmental assessment	7
Auditory processing condition (APD) and audiology	5
Normed based general assessment (e.g. BASCIII, ECBI, CBCL)	4
Physical and medical assessment	4
Review of Cognitive assessment	4
Sleep review	3
Occupational therapist assessment	2
Learning report (Educational psychologist, teacher or RTLB)	2
Other assessments guided by findings	6
Possible diagnoses	
ADHD	23
Autism	12
Sleep condition	7
Specific learning disability	5
Auditory processing condition	4
Trauma	3
Oppositional defiance condition	2
Attachment	2

Sensory processing condition	2
Foetal alcohol spectrum condition (FASD)	1
Anxiety	1
Tourettes	1
Etc. (e.g. more diagnoses considered depending on information)	8
Possible Services/Interventions	
Resource teacher for learning and behaviour (RTLB)	11
Medication (if indicated by ADHD assessment)	6
Parent education (Parent courses including Triple P parenting)	6
Psychology referral	6
Support in school setting	5
Sleep programme	5
Special enducation needs co-ordinator (Senco)	5
Behavioural intervention	4
Social skills training continued	4
Information shared with parents	2
Learning support co-ordinator	2
Not applicable	2

Possible Diagnoses

Table 6.5 indicates that clinician considered different possible diagnoses for the 8-year-old with the most common being ADHD.

Recommended Services

Table 6.5 indicates that the clinicians would recommend 11 different services based on the vignette. The average number of services recommended by each clinician was two. The most common was a referral to a resource teacher for learning and behaviour (RTLB), that is, an experienced teachers who support children in an educational context. The majority of the services involved a referral to another programme or colleague specialty such as specialist teachers, called resource teachers for learning and behaviour (RTLB) in New Zealand.

Discussion

In this study, clinicians indicated that a vignette of a complex case would lead them to consider a number of possible diagnoses, assessments, and support services for children both 4-and 8-years of age. The response to the vignettes corroborate previous research that referral descriptions of multiple neurodevelopmental conditions suggests case-complexity and do not result in a universal pathway to assessment and management (Coughlan et al., 2022; Male et al., 2020). The pressure to diagnose neurodevelopmental conditions quickly and accurately stems from the mounting evidence for impact of early diagnosis and support on life outcomes (Whitehouse et al., 2021). The current study suggests that complex cases may be more difficult to diagnose with particular described characteristics, not indicated in neurodevelopmental diagnostic criteria, but of important clinical concern.

The clinicians showed overall lower levels of concern for the characteristics of the four-year-old child than that described for the 8 year-old-child. This might be due to variations in developmental expectations of behaviour, such that "roaring loudly" is appropriate for a four year-old, but difficulty appropriately modulating volume for eight-year-old is of greater concern (Halperin & Marks, 2019). However, the greatest concern for the four-year old child was elopement. Elopement or wondering, which is defined as leaving an area without permission and entering a potentially dangerous situation, is exhibited by a third to half of autistic children and causes immense stress for families (Anderson et al., 2012; Pereira-Smith et al., 2019). Elopement is not part of autism diagnosis criteria, however as a clinical concern, it is the most likely of all the described characteristics to potentially cause physical harm to the child (McLaughlin et al., 2020). Elopement, as described in the vignette was placed as an example the DSM 5 ADHD characteristic of 2c) often runs about or climbs in situations where it is inappropriate, but it is possible that the higher level of concern is not that of an ADHD characteristic, but the issues of safety and wellbeing. Equally, 'difficulty

sleeping' was a relatively high level concern for clinicians with the eight-year-old description, but this issue is not part of a diagnostic criteria, so much as a concern for the well-being and development of the child (Martin et al., 2019).

The services suggested by clinicians also highlight another challenge for practitioners, which is that of readily available and immediate support for families. In general, the suggested services involved referrals on to other organisations and colleagues. Research suggests that families have an expectation that clinicians will give immediate help and access to services, however, in many countries, including New Zealand, clinicians refer to other specialty services for support (Kasilingam et al., 2021). This is often the part of the process that families are most dissatisfied with, where immediate support is seemingly out of reach or requires more waiting and "qualifying" (Daniels et al., 2021; Makino et al., 2021; Ooi et al., 2016).

Clinicians are in demand in New Zealand with often long waitlists (Eggleston et al., 2019). This stretched-work-force is under time pressure, and therefore it is possible that qualitative answers did not represent the full breadth of knowledge and options that would be considered. This also meant that the sample size was small for this study and statistical analysis was not possible. Furthermore, the questions in this survey asked clinicians to base their self-reported answers on the vignette when clinicians often gather more information in practice leading to a next line of assessment, intervention or hypothesis. Therefore, a limitation is that this is not representational of complete practice. As the vignettes were designed to demonstrate real-world diagnostic complexity, this meant that it was not possible to directly compare the 4- and 8-year-old vignettes.

The number of assessments and possible diagnoses that the clinician considered in the current study suggests that considerable resources are required to adequately diagnose complex cases. In the referrals that clinicians receive, the greatest concerns may not be those

that might indicate a clinical diagnosis so much as those concerns that indicate possible safety or well-being issues. Greater communication and clarity around the important concerns, possible number of assessments needed to provide an accurate diagnosis, and necessity to refer for specialised support, might improve communication around the diagnostic pathway.

CHAPTER 7

GENERAL DISCUSSION

A biopsychosocial model is a wholistic model that considers a number of relationships, and interactions are necessary to understand issues in education, health and well-being (Lehman et al., 2017). These models, like Bronfenbrenner's bio-ecological systems theories (Bronfenbrenner, 1992), and indigenous health models like Durie's Te Whare Tapa Wha (Durie, 2004) state that studying phenomena in a biomedical paradigm is limited. Therefore, this thesis examined the diagnosis of autistic and ADHD children from multiple perspectives following a biopsychosocial model. This was achieved by considering contributing factors from a biological and development perspective and accounting for age and atypical development. In addition, more psychological and social factors were considered, such as the possible thinking process of clinicians when presented with complex cases and also social and psychological perspectives of parents, who experience diagnosis for their child.

This biopsychosocial model means that the answer to the question of what causes the delay in autism diagnosis with ADHD co-occurring is not just less parent reported atypical development during the first year of life, but also parents' perspective that the system is difficult to negotiate, and that co-occurrence is complex to diagnose, meaning increased time and resources. This model has aided the interpretation of results, by not only ensuring that the biological, social and psychological aspects of the diagnosis process for multiple stakeholders is considered, but that the interactions between these aspects and stakeholders are also a focus. The relationship dyad between clinician and parent is of particular note, because the social and psychological positioning of each seems to be misunderstood. Delays in diagnosis are conceived by parents as due to the system (waitlists, parenting courses, lack of transparency around the process) or a particular clinicians' beliefs. This appears to diverge

from a clinicians' need for time to understand multifaceted biological factors and conduct a thorough assessment. In complex cases, this might include using a range of general and specific diagnostic assessments to afford sufficient data to support or discount diagnosis, and adherence to best practice that considers clinical thresholds. For the majority of both parents and clinicians, their purpose appears to be the same in helping the child to get the correct diagnosis in order to access support or treatment. Therefore, a biopsychosocial model illuminates these interactions, with the possibility that greater communication and understanding around the positioning of parents and clinician might reduce wait times and, ultimately, delayed access to support for the child and their whanau.

The weight of each factor to cause a delayed autism diagnosis with co-occurring ADHD is beyond the scope of this research, and it is likely that each individual circumstance of diagnosis has different weight on these factors. Participants were informed of the results of the studies via a user-friendly "research snapshot," which was shared with Autism NZ, ADHD NZ, or provided by request from participants [see Appendix G].

Therefore, returning to the questions outlined at the outset of the this thesis, the first question asked whether there is evidence in New Zealand for differences in the timing of diagnosis when autism + ADHD co-occurs compared to populations with only one of those diagnoses. The second question extended this line of investigation by asking what factors might be influencing the age of ADHD and autism diagnosis. This involved a number of sub-questions that tested possible hypotheses for the delay, such as does parent reported atypical development influence age of diagnosis, do certain presenting concerns to a health professional trigger a quicker diagnosis pathway, or are there other delays, such as the timing of seeking specialist help or the number of types of specialists seen prior to diagnosis? The third question probed similarities and contrasts in the diagnostic patterns across the group by

asking if children with autism + ADHD resemble an early trajectory of development and help-seeking of an ADHD only or an autism group or their own unique pattern.

The fourth question focussed on the experience of diagnosis as reported by parents of the three diagnostic groups. This question asked what parents' perception of the diagnosis process added to our understanding of the experience and possible delays. In adhering to a broader examination of the stakeholders involved as befitting a biopsychosocial model, the final two questions focused on the role of clinicians. The last two questions asked what would be the assessments, hypothetical diagnoses, and interventions clinicians might recommend for complex co-occurring neurodevelopmental cases and is there a consistent pattern or pathway recommended for complex co-occurring neurodevelopmental cases?

The above questions are summarised below:

- Q1. Is there evidence in New Zealand for timing differences in diagnosis when autism + ADHD co-occurs compared to a populations with only one of those diagnoses?
- Q2. What factors might influence the age of ADHD and autism diagnosis?
- Q3. Are children with autism + ADHD similar or different in their developmental trajectory and help-seeking behaviour than children with ADHD only or autism only?
- Q4. What can parents' perception of the diagnosis process for either autism, ADHD or autism + ADHD add to our understanding of the experience and possible delays?
- Q5. What would be the assessments, hypothetical diagnoses, and support services clinicians recommend for complex co-occurring neurodevelopmental cases?
- Q6. Is there a consistent pattern or pathway recommended for complex co-occurring neurodevelopmental cases?

Main Findings

International research suggested a particular phenomenon in the interaction between two co-occurring conditions, whereby when ADHD is present with autism, an autism

diagnosis is given significantly later than an autism only diagnosis (Jónsdóttir et al., 2011; Levy et al., 2010). The literature review (Chapter 2) further established the existence of this phenomenon and also elucidated that an ADHD diagnosis is given significantly earlier when autism is present, compared with an ADHD only population.

The three studies in this thesis sought to analyse and understand these differences in diagnostic timing and process from a number of perspectives. The first aim was to determine that the delay in autism diagnosis when ADHD was present occurred in a New Zealand population. A sample of 288 New Zealand parents reported a median delay of 3.2 years for an autism diagnosis when ADHD co-occurs compared to an autism only population (Q1). There was also evidence that an ADHD diagnosis occurs 1.4 years earlier for the co-occurring autism + ADHD group compared to an ADHD only population (Q1).

After establishing evidence for this delay, quantitative analysis established that autistic + ADHD children had different atypical development from autistic only children in the first year of life, with parents reporting less language and social issues, but increased stereotyped behaviour at 7-12 months of age (Q2). Autistic + ADHD children sought later specialist help and saw a greater range of types of specialists than autistic children. The nature of the concerns presented to specialists were also less likely to be symptoms of autism or ADHD (Q2). Other more general patterns in diagnosis included that referral concerns that indicated a particular symptom of autism or ADHD were associated with a faster time to diagnosis. Over 40% of the ADHD and co-occurring group presented with "other" characteristics, meaning other behaviours outside of a diagnostic criteria. The different diagnosis groups had quantifiably different patterns in atypical development, early help-seeking, presentation concerns, wait times and number of types of specialists encountered (Q3). This suggests that these factors might contribute to explaining the different timings in diagnosis.

It was expected that there might be some parents who preferred an alternative explanation to their child's current diagnosis. However, it was not hypothesised that each group would have similar rates of one fifth of parents stating an alternate option within the three diagnoses would be more suitable for their child. It was also hypothesised that if a parent did believe an alternate diagnosis to be more appropriate that it might sit outside the triad of autism, ADHD or autism + ADHD; however, this was not the case. It is not clear as to why this pattern was found, but from a gradient hypothesis theory of autism + ADHD co-occurrence, this might be taken as evidence of the continuously distributed characteristics of autism and ADHD (van der Meer et al., 2012).

The results of reflexive thematic analysis from Study 2 suggest that parents generally perceived the diagnosis process for autism, ADHD, and autism + ADHD as a negative adversarial experience where delays and inequities come to be expected (Q4). Parents, for the most part, did not perceive a collaborative, helpful system. Instead parents reported that they are isolated as advocates for their children. Their identity as parents and the child's identity are also negatively impacted during the diagnostic process, but this negativity is usually replaced by a positive reframing once the diagnosis is received. Parents express that there are multiple criteria their families must fit to access diagnosis and support. In particular, generalised parenting courses are viewed as an unhelpful tool, especially when used as a perceived gatekeeper to diagnosis.

Parents' perceptions of the identification to diagnosis process for their children diagnosed with autism, ADHD, autism + ADHD differed surprisingly little across the three diagnosis categories (Q4). Key perceptions for the autism groups was a desire for dialogue about neurodiversity to occur earlier in the diagnostic process. Within the ADHD diagnosis, there was greater focus on the role of medication and the challenges with ongoing

monitoring, as well as the transformative effects of medication. The ADHD groups also encountered the age criteria of being told they were "too young" for a diagnosis.

In Study 3, clinicians recommended a diverse range of assessments, hypothetical diagnoses, and interventions in relation to the hypothetical cases (Q5). Suggested assessments were specific to particular diagnoses, but sat alongside wider developmental, and more general, assessments. Most clinicians recommended immediate assessment. There was no consistent pattern or pathway recommended for the described complex co-occurring neurodevelopmental cases (Q6), however the process by which clinicians indicated they might approach a complex neurodevelopmental case generally lengthy and resource heavy. Some characteristics described in the vignettes were of greater concern than others due to issue around the child's safety or well-being. Clinicians prioritised these concerns over characteristics that might fit diagnosis criteria. The hypothetical cases were designed to be indicative of case complexity, and the results imply that such cases with co-occurring issues require a different level of resourcing, or possibly triaging, which sits outside of singular case presentation.

Quantitative data from Study 1 and 3 and qualitative data from Study 2 indicate factors that might be contributing to a delayed autism diagnosis with co-occurring ADHD. The additive effect of these two diagnoses means that autism + ADHD are complex cases to diagnose. The complicating factors include, a blending of "symptoms" of both diagnoses and other atypical characteristics, a different presentation pattern, and atypical development from either singular diagnosis. It is difficult to speculate on cause and effect, but these factors possibly contribute to what is an already lengthy and opaque diagnostic process. Clinicians are also in a difficult epistemological position of trying to ensure co-occurrence is accounted for with a concern not to over or under diagnose. Study 1 demonstrated this with the number of types of specialists seen for the co-occurring group, and different atypical development

and presentation patterns from the groups with singular diagnoses. This was also described in the voices of parents in Study 2, who lamented the wait times, the difficulty of navigating the diagnostic system and also the difficulty of meeting diagnostic criteria thresholds at particular time points and in short clinic visits. Finally, Study 3 showed that clinicians use a number of different assessments for complex cases and prioritise characteristics that imply safety or well-being concerns. This suggests that there is lengthy time and resources involved in diagnosing complex cases, which involve co-occurring neurodevelopmental diagnoses and other possible factors. These studies are timely as co-occurrence rates increase, and there is a growing concern to diagnose promptly and accurately in order to access early supports (Soke et al., 2018; Waddington et al., 2021; Whitehouse et al., 2021).

Contributions to the Research Literature

Due to the hierarchical specifications that were in place in the DSM until 2013 which prevented a clinical recognition of co-occurring autism + ADHD (Miodovnik et al., 2015), this thesis contributes to an understudied area. The literature review mapped the wide geographical prevalence of the delay in autism diagnosis when ADHD is present. Although a number of studies had suggested this was a common pattern (Jónsdóttir et al., 2011; Levy et al., 2010; Miodovnik et al., 2015), the literature review contributed a synthesis of this phenomena across multiple studies and geographical locations. The literature review collated the information to provide evidence of an international pattern.

The review also indicated a number of other factors that appeared to impact a co-occurring group's pathway to diagnosis. These factors included parents having later concerns and waiting longer for a diagnosis. The review also highlighted that this delay could be compounded by certain features which appeared to add to the presentation of case complexity. These included being female and being diagnosed with ADHD first. When ADHD was diagnosed first, the implication was that the delay in autism diagnosis was

exacerbated, and researchers have hypothesised that ADHD was more apparent during short clinic visits and that a number of active and passive processes for clinicians might be occurring (Heyman et al., 2022; Miodovnik et al., 2015). The process included "search satisfying," overshadowing or masking of the autism traits by the ADHD symptoms, or substituting ADHD for autism + ADHD due to a greater familiarity with ADHD diagnosis process (Miodovnik et al., 2015; Soke et al., 2018; Stevens et al., 2016; Wallisch et al., 2020). There was inconsistent data on the effects of intellectual difficulty when coupled with autism + ADHD and the effects on diagnosis (Stevens et al., 2016) .

Only three studies compared the timing of an ADHD diagnosis between an ADHD only group and a autism + ADHD group (Engelhard et al., 2020; Jensen & Steinhausen, 2015; Lin et al., 2014). All three studies recorded an earlier diagnosis of ADHD when autism is present (Engelhard et al., 2020; Jensen & Steinhausen, 2015; Lin et al., 2014). The 12-20 month earlier diagnosis of ADHD when autism is present compared to an ADHD only population, often passes without comment because it an expected pattern of diagnosis. That is, it appears logical that greater severity would lead to earlier help-seeking (Engelhard et al., 2020; Jensen & Steinhausen, 2015; Lin et al., 2014). However, this finding only makes sense when not contextualised in relation to the autism only timing of diagnosis. In that, the same logic should follow that this "severity" would also lead to earlier help-seeking and therefore, an autism diagnosis for an autism + ADHD child being given at the same time, or earlier than an autism only population. Therefore, while more characteristics than an ADHD only population might lead to earlier specialist help and, thus, an earlier diagnosis compared to an ADHD only population, other factors might be contributing to a delayed diagnosis of autism. However, the results of Study 2 suggest no significant difference between an ADHD and autism + ADHD population in the age at which specialist help was sought, but again a significant difference in the early ADHD diagnosis of the autism + ADHD population. The

explanation for this is not clear. One possible contributing factor might be evident from the qualitative study where parents of ADHD only children mentioned clinicians preferring to wait until their children were six years old before giving the diagnosis of ADHD. The children with autism + ADHD might be more likely to be given the ADHD diagnosis at the same age as their autism diagnosis or, perhaps "severity" leading to a softening of the recommended age for diagnosis of ADHD.

These studies from a large New Zealand sample have also contributed to the research literature by demonstrating, compared to a single diagnosis, there is a delay in autism diagnosis when ADHD is present and that ADHD is also diagnosed earlier when autism is present. Beyond adding another country to the list of where this phenomena occurs, this research has provided further and more specific evidence for where along the diagnosis pathway this delay might be occurring. Specifically, the autism + ADHD group had different parent reported patterns of atypical development possibly leading to later specialist help-seeking. In addition, parents of autistic + ADHD children appeared to present to specialists with a concern that was less likely to be an autistic-characteristic, leading to more types of specialists seen and a later diagnosis compared with an autism only group. This level of detail and examination in relation to the delay of diagnosis had not previously been studied across the three diagnoses.

The role of observed atypical behaviour during the first year of life is a burgeoning area of research (Guinchat et al., 2012; Waddington et al., 2022). The relationship between the number of observed areas of atypical development and atypical development in areas pertaining to autism diagnosis criteria have been shown to lead to earlier help-seeking and earlier diagnosis, enhancing the potential for early support (Guinchat et al., 2012; Waddington et al., 2022). However, the findings of this thesis provide an understanding of what these patterns of observed areas of atypical development during the first year of life

might look like across the three different diagnoses. The results of the Study 1 suggest that in social behaviour, language, and atypical motor development clusters, the autism group showed the highest parent report of atypical behaviour at 6-12 months, while autism + ADHD had lower rates of atypical development in these areas. In the stereotyped and restrictive and behaviour/temperament cluster, the trend was reversed with autism + ADHD group being higher and the autism group lower. However, in most cases the ADHD group had significant differences and reported half or less than half the rates of atypical development in these areas. The exception for the ADHD group was the behaviour/temperament domain.

The ADHD group showed similar atypical development to the autism groups in sleeping difficulties, medical issues and behaviour/temperament. These patterns of development contribute to the research literature a greater understanding of early parent reported atypical behaviour specific to diagnoses and indicate possible reasons for delays in diagnosis. The research suggests that the greater overall amount of reported atypical development for the autistic group during the first year of life contributes to an earlier diagnosis (Guinchat et al., 2012; Waddington et al., 2022). There is also a suggestion that significantly higher areas of atypical development, which fit less of a diagnostic-specific criteria, such as sleep and behaviour and temperament issues, might contribute to a more complex-case paradigm, and thus a delay in diagnosis. These areas, which are less-diagnosis specific, were particularly high areas of atypical development reported by the ADHD and autism + ADHD group compared to other areas of atypical development that can lead to faster diagnosis times, such as concerns around language and communication (Guinchat et al., 2012; Waddington et al., 2022).

Another contribution to the literature is a greater understanding of the relationship between the primary presenting concern that a parent describes in interaction with a specialist

and time to diagnosis. In our sample, a presenting concern which has diagnostic ambivalence, such as behavioural difficulties, led to a longer time to diagnosis. The autism group demonstrated remarkable consistency in presenting with a concern that fit with an autism diagnosis criteria. The autism + ADHD group and ADHD group had over two fifths of parents present with "other" concerns. Examples of these 'other' concerns included descriptions of "aggression/violence", "emotional dysregulation", bullying, and learning difficulties. Both autism + ADHD and ADHD group had significantly longer wait time to diagnosis than the autism group. This contributes to the literature by suggesting a possible relationship between presenting concern and wait times to diagnosis. It also suggests that the ADHD and co-occurring group are more similar in this pattern than the autism only group.

Eggleston et al. (2019)'s study of parents' experiences of their child receiving an autism diagnosis found that co-occurring ADHD was predictive of a higher number of professionals consulted prior to diagnosis. However, Study 1 added that there was a significant increase in the number of different types of specialists seen compared to the autism only group. The ADHD group had no significant difference compared to the co-occurring group in terms of the number of *types* of specialists seen. There were quantifiably more significant parent observed areas of atypical development across almost all areas for the co-occurring group compared to the ADHD only group. Thus, it is possible that quantity and higher need plays a role in accelerating an ADHD diagnosis, but it was hypothesised that this would result in an earlier age at which specialist help was needed compared to the ADHD group. This was not the case.

Parents' experience of the autism diagnosis has been well documented in the research literature (Legg & Tickle, 2019; Milner et al., 2019), however, the studies in this thesis enabled the examination of similarities and differences across these three groups. The results suggest multiple similarities in the experience of the diagnostic process across different

neurodevelopmental conditions. In particular, parents' experience of the ADHD diagnosis has not been well documented with emphasis instead on the experience of living with an ADHD child and effects of medication (Carr-Fanning & Mc Guckin, 2018; Silva et al., 2015). In addition, the diagnostic experience of co-occurring autism + ADHD has not been examined, with more of a focus on parental stress with either diagnosis (Martin et al., 2019; Ooi et al., 2016). The contrasts in themes also highlighted key contributions to research literature, such as the burgeoning positive emphasis on the neurodivergence in the autism community that is not currently apparent in an ADHD community. The thematic analysis also showed how parents linked the diagnosis experience to their identity as a parent, which shaped their reactions to situations, such as parenting course-supports.

The second study employed a relatively novel use of reflexive thematic analysis and followed other reflexive-thematic models of creating a thematic map after dwelling with the data (Terry et al., 2018). The use was novel as there is some debate around whether the data retrieved from an online survey is rich enough to warrant a reflexive analysis, and arguably, this was compounded by the survey including quantitative questions (Braun et al., 2020). However, the richness of the data was demonstrated in analysis, and the importance of the reflexive thematic themes contributes to the growing literature using this methodology and the possibilities of mixed methods and online tools being used in reflexive thematic analysis. Thematic analysis in psychological research usually follows the six step process outlined by Braun and Clarke (2006). Recently, this process was updated to include different steps and a call to acknowledge the subjectivity of the researcher, the ineffectiveness of interrater reliability and to encourage "dwelling with the data" (Braun & Clarke, 2021). Reflexive thematic analysis was also critical of certain devices used to organise themes, such as barriers and facilitators, arguing that this is too superficial for a reflective thematic analysis (Braun & Clarke, 2021).

The third study also involved a relatively novel vignette-based methodology to explore the clinicians' recommendations for complex co-occurring neurodevelopmental cases. A vignette methodology has been used in ADHD research to examine teachers' perception of inattentive behaviours and possible supports in the classroom (Groenewald et al., 2009; Moldavsky et al., 2013) and parents' perception of ADHD behaviour challenges often with a male and female vignette (Moldavsky et al., 2013). The advantage of a vignette case referral description used in the third study of this thesis is that it did not dictate a set diagnosis to clinicians, such as asking how they might assess for autism + ADHD, but instead, attempted to present a more naturalistic complex case, which did not presume a correct answer. This methodology thus sought to understand the range of possible approaches that clinicians might take to complex-cases (Evans et al., 2015). The thesis contributes to the research literature by using this methodology to show that there is no set pattern to assessing complex co-occurring cases but rather, clinicians use a diverse range of assessments and referrals. The range of multiple assessments and referrals also suggests extensive resources and time are needed to assess case complexity.

In conclusion this thesis has contributed to the research literature by adding to our understanding of the reasons why complex cases might receive a delay of a co-occurring diagnosis. The quantitative results suggest the delay in receiving an autism diagnosis with co-occurring ADHD is caused by more complex case presentation to health professionals and less key atypical development recognised as part of autism criteria compared to an autism only group. The qualitative data builds on this by exploring parents' perspectives on the adversarial diagnostic process. Parents view referrals and parenting courses, not as thorough or best practice, but as unnecessary delays. The final study demonstrated that clinicians approach complexity with both specificity and more general assessments, requiring multiple

considerations and assessments. This suggests that these complex cases require more time and resources than cases which appear to fit with diagnostic criteria.

Implications

There are several important implications arising from this thesis. First, this thesis suggests that parents of autism + ADHD children may report similar atypical development to children diagnosed with autism alone. However, some key areas of differences might help clinicians be alert to co-occurring diagnoses. The pattern of a child with both autism + ADHD presenting with less language and social concerns, but more stereotyped behaviour, might alert clinicians to consider an autism diagnosis. The possibility that autistic + ADHD children often present with later and less specific concerns to a specialist can also help the diagnosis process by again alerting clinicians and parents to these patterns. It is by understanding these different patterns of behaviour and observation that clinicians, policy makers and parents can ameliorate delays in diagnosis.

The research also has implications for children diagnosed with ADHD. There is a wide variety in expectation of onset of ADHD traits between 2.5 and 7.5 years and ADHD diagnosis occurring anywhere between the ages of 6.2 and 18.1 years (Rocco et al., 2021). However, the implications of the study on atypical development during the first year of life indicates that, although significantly less than the autism groups, there are unique patterns to development which can lead to earlier diagnosis. If clinicians understand that marked sleeping issues and behaviour and temperament concerns during the first year of life is similar or higher than an autism group, this might flag a possible consideration of ADHD. This in turn might allow for access to early support.

In addition, specialists might consider that a first diagnosis of ADHD should not preclude further assessment of autism, or that autism should at least not be ruled out. Specialists might also want to consider discussing with parents, whose children are first

diagnosed with ADHD, the key symptoms of autism so that parents feel empowered to return for the possibility of an autism diagnosis sooner or consider explaining that the first diagnosis is not necessarily a diagnostic endpoint. These suggestions have the potential to reduce the delay in diagnosis for autism + ADHD children by helping ensure that parents report autistic characteristics sooner, and improve the diagnosis and treatment for children and their families.

Parents' qualitative descriptions of their experiences of an ADHD diagnosis suggest that a possible cause of an earlier ADHD diagnosis for children co-occurring diagnoses might be that parents of ADHD-only children are actively advised to wait until their child is over six years in line with recommendations around the importance of the school environment (Halperin & Marks, 2019). The implication is that all ADHD children get to the starting line of diagnosis (seeking specialist help) on average at the same time, but that the ADHD only children are held back from finishing, while the increased atypical development of the co-occurring group, and the pressure from the autistic and research community to diagnose autism earlier results in the ADHD diagnosis being given sooner.

The relationship between the nature of the presenting concern to a health professional and time to diagnosis has multiple implications. One implication is that parents might consider emphasising certain concerns that trigger diagnosing criteria in order to accelerate their access along the diagnosis pathway. Clinicians might consider how ambiguous concerns might indicate co-occurrence and complexity and choose to accelerate access to multi-disciplinary diagnostic teams to ameliorate a delay that occurs in complex cases. The delay in help seeking for these groups might be due, in part, to the nature of the child behaviour in that it might be attributed to temperament, behavioural difficulties or parenting rather than requiring specialist intervention (Wallisch et al., 2020; Yamauchi et al., 2015). The

implication might be that behavioural concerns possibly attributed to parenting might be better triaged as indicative of a complex case requiring referral to a multidisciplinary team.

The implications of a fifth of parents in each group believing the co-occurring category a better fit than either singular diagnoses and a fifth of the co-occurring group parents favouring a singular diagnosis are unclear. This might be used as evidence for the overlap of the diagnoses or the gradient model of autism and ADHD as indicated by Van der Mer et al. (2012). Arguably it might be a study bias whereby autism and ADHD were subconsciously on parents' minds as alternative options. However, it does suggest that within each diagnostic category there are a proportion of parents who believe that the diagnosis is more accurately described by an alternative combination of these three groups. This might also indicate an expected fluidity between the diagnostic groups by which one fifth of the singular diagnosis groups might expect to move into the co-occurring diagnosis group in the future.

A reconciling of the parent and clinician positions through greater communication of the diagnosis process and expectations might strengthen these encounters and create a stronger sense of collaborative purpose. This collaboration might include communication and clarity around: the difficulty of the diagnosis process, particularly for complex cases; the length of the required assessments; the possibility of co-occurrence; and the availability of support after diagnosis. In addition, an openness to validate the parents' position as a "good enough" parent (Winnicott, 2021) with a neuro-divergent child while also being aware that adopting a positivist position might encourage a sharing of family trauma and other difficulties, that might also inform the diagnosis process. Clinicians might also communicate that some delays in diagnosis are not due to the system or particular clinicians' beliefs, but rather the complexity of a case. The lengthy time taken is often clinicians adhering to best practice, by triangulating data and using multiple assessments, to consider wider possibilities,

co-occurrence, and specific diagnoses. This understanding would help manage parent expectations that diagnosis of complex cases is a quick process.

This research has demonstrated many reasons for the pursuit of a label that have been echoed in other research. These include the autism label as a positive identity, with a sense of belonging to a community (Krcek, 2013), the label as resituating feelings of guilt or blame (Daniels et al., 2021), and the label as means to access support (Ooi et al., 2016). Parents often want these neurodevelopmental labels, sometimes consciously or subconsciously, for some or all of these reasons (Daniels et al., 2021; Krcek, 2013; Ooi et al., 2016). This parental-position, epistemologically challenges the discipline of educational psychology in New Zealand, because there has been a paradigm shift to support children and parents, which involves moving away from diagnostic bio-medical model to using a more ecological approach that focuses on changing the environment around the child (Gutkin, 2012). It is possible to hold both paradigms, where by an ecological approach is adopted after the child's diagnosis to better adapt the environment, based on the child's need and other evidence-supports for that diagnosis. However, the difficulty comes when parents encounter professionals, along the diagnostic pathway, rather than afterwards, who they believe will expediate a diagnosis, but adhere to a different paradigm approach. The bio-medical-diagnosis model is less than ideal, but without alternatives which are able to achieve the shifts in identity, thinking and sense of community, professionals might wish to reconsider the way that their paradigm and scope of practice reflect the needs of a family, who are moving towards a diagnostic-model.

Parents might also view the diagnostic process more positively if clinicians could provide access to immediate interventions, such as behavioural support for the child and counselling for the parents. If clinicians were able to access services, this might further remove the time pressure to accurately diagnose complex-neurodevelopmental cases.

Although parenting-courses might be considered an immediate support which parents can access, greater sensitivity to how these supports might be interpreted by parents would appear to be beneficial to parent identity, engagement and well-being.

There are also implications for professionals working to diagnose complex cases. The complexity of presentations means that a two level triage system may be beneficial (Whitehouse et al., 2018). This would involve cases, which appear to be more straight forward to diagnose, being seen by a singular clinician and more complex cases being accelerated to a multidisciplinary team, with the knowledge that the latter cases will take longer and require more resources. The research also suggests that greater support of clinicians, who need to undertake often lengthy and resource heavy processes when they consider diagnosis of complex cases, is warranted.

Limitations

A limitation of this thesis is that autism + ADHD have only recently been permitted to be diagnosed together under the DSM 5 (American Psychiatric Association, 2013). A population between 0-18 across all studies means that some children were diagnosed under the previous specifications, which had a hierarchal diagnosis structure that premised that autism diagnosis would be given only, even if ADHD symptoms were significant. This means that the diagnostic groups that involved children diagnosed prior to 2013 may not be representational of the true prevalence of co-occurrence. It is also possible that the current co-occurrence group is at the higher support needs end of the spectrum for both diagnoses due to some of the group obtaining a diagnosis during a period of time when it was not universally recommended. However, Goldstein and Schwebach (2004)’s study, which reviewed clinical charts, points out that clinicians had an obligation to diagnose autism + ADHD before 2013, particularly when treatment for ADHD was warranted. Nonetheless, a limitation remains that

the data used in these studies might be limited by the change in position reflected in the DSM-5.

A further limitation might be that the study should have included adults who had a late diagnosis. This is an expanding area of research with increased numbers of adults being recognised as having autism and/or ADHD (Fusar-Poli et al., 2020; Lai & Baron-Cohen, 2015; Mucci et al., 2018). The specifications of diagnostic criteria state that the characteristics of both ADHD and autism must be present in early childhood, so an adult diagnosis is a missed or late diagnosis, although there is a debate about whether late onset is possible (Mucci et al., 2018). This population might be able to give first-hand experience of the diagnosis process and add to the understanding of timing around these diagnoses.

Sample size is a limitation across the studies While the overall number of participants in the survey was large (n=288) this is reduced when divided into diagnostic groups. Greater participant numbers, particularly looking at the prevalence of a phenomena such as delayed diagnosis, could further strengthen the results (Trotter, 2012). This limitation of sample size was particularly acute in the final study of clinicians, across multiple disciplines. The small number of clinicians cannot be seen to represent the clinicians of New Zealand. Sample size is also debated across quantitative and qualitative data sets. For the qualitative data set, further saturation through additional interviews and focus groups to ensure themes were exhausted, could have further strengthened the qualitative analysis (Trotter, 2012). There are also inherent limitations to online surveys (Andrade, 2020). Two limitations include; it is difficult to describe, or delineate, the exact population who answered, and participants with particular biases might self-select to answer (Andrade, 2020). An example of participants self-selecting might be that parents participated if they had experienced a predominantly negative experience with their child's diagnosis.

A further limitation is the time the participants were able to give to answering the survey. Time limitations have the potential to restrict participation in the survey or significantly curtail participants' responses. This limitation was expressed particularly in the clinicians' survey. Clinicians wrote in short-hand that multiple other things would be considered, such as further assessments informed by a step-one approach, and that in some cases, they would not speculate a diagnosis without first having more information.

The methodology of Study 1 and Study 2 required retrospective recall. The limitation of this methodology is the fallibility of memory. The memory of the diagnosis process might also have been different across the different age brackets, with parents who experienced diagnosis a decade ago encountering a different system and remembering a more flattering experience. Study 3 required hypothetical-assessment-plans based on a small amount of referral information and it is possible that there was also a bias to portray an idealised response to a hypothetical case rather than a real-life representation of the clinicians' process.

Furthermore, the methodology of all the studies aimed to incorporate a biopsychosocial perspective on diagnosis, but this will be invariably limited in that there are so many multiple perspectives and viewpoints along a diagnostic pathway (Tramonti et al., 2021). This was also not a direct-observational methodology and relied on parents observation of their children, which created a limitation in that the researcher was using a third-party report of a phenomena (Tramonti et al., 2021). This thesis was conceived to capture the parents' experience of diagnosis across three different diagnostic groups in order to better understand the timing of diagnosis. Research suggested that the role of clinicians in this picture was also important, however there are a number of other systems and stakeholders, who experience has only superficially been touched on. These groups include fathers and caregivers due to the limited participation in the survey, but also the teacher's role and the voices of children. The voices of teachers, children, clinicians and fathers were

limited in contribution to this biopsychosocial perspective on the diagnosis of autistic, and/or ADHD children.

Future Research

The complexity and individual needs of neurodivergent children are increasingly being conceptualised by co-occurring labels, therefore understanding and studying co-occurrence is vital. Valderas et al. (2009) biomedical example of framing co-occurrence remains a key philosophical underpinning, but greater theory looking at how co-occurrence might construct a neuro-divergent identity and, greater understanding for the experiences of co-occurrence for children and family, is needed. This might include a study on the relationship of identity formation when a child has co-occurring diagnosed health needs. For example, research could focus on the terminology preferences for people with co-occurring diagnoses.

One of the implications of this thesis is a suggestion that clinicians consider sharing with parents the difficulties of diagnosing complex cases, the lack of stability of neurodevelopmental diagnosis, and the possibility of co-occurrence. The criticism of such a position, is that it would undermine the credibility of the clinician (Daniels et al., 2021), therefore future research might consider testing if this is the effect of more transparent collaboration. This research might include asking parents at what stage and if that information would have been useful and also asking clinicians for their input around potential change. The next step might be considering a systematic implementation of a change in a pilot sample and examining what clinicians and parents think.

Another key area of potential research is greater co-design with the autistic + ADHD community to understand what is the lived diagnostic and support experience of co-occurrence and what might enhance autistic + ADHD quality of life (Monk et al., 2022). Future research might consider if, and when, an autistic person gained a sense self-advocacy

and knowledge of neurodivergence, and whether this was within a diagnostic system, or after the label was given (Monk et al., 2022). Furthermore, understanding the role of co-occurrence within self-advocacy and concepts of added gifts, or exceptionality, with additional neurological differences, might help promote greater advocacy for the co-occurring community.

One way that the current research differed from most previous studies was that there was an implied social shift in parents desiring a label for their child. This was suggested in the reflexive thematic analysis, where parents described aiming for a label/diagnosis at the outset, because it was believed that this would ensure access and support and relieve the "blame." Future research might consider exploring if there is evidence for this social shift on a broader level. Understanding parents and teachers positioning around labels and access to intervention might help strengthen an understanding of the motivation to diagnosis and if access to supports was equally offered without a label, whether there would be a significant decrease in motivation for a diagnosis.

The focus of this thesis has been on the diagnostic pathway for families and clinicians to understand the delay of an autism diagnosis when co-occurring ADHD is present. Future research might consider a greater understanding of response to intervention and the best ways to tailor supports for an autism + ADHD group. Intervention outcomes for autistic + ADHD children are reduced in effectiveness, such as social skills training being less effective with autistic children who also have ADHD (Flouri et al., 2015). Future research might consider understanding how effective support for autism + ADHD can be adjusted or more generally, how interventions can address neuro-complexity.

The relationship between diagnosis, label and support or access to intervention deserves to be further elucidated. The crux of parental frustration about the delay to diagnosis is the implication that their child remains in limbo for this period of time without access to

support and understanding during crucial developmental years. We have societal concerns about increased diagnosis and medication of children (Sayal et al., 2018), but this appears to be philosophical, or even, insensitive when a parent is trying to access a system to help their child. Future research might consider how to bridge this gap. This might include a study of neurotypical communities' attitudes to motivations for diagnosis, neurodiversity, medication and support.

It is difficult to know how to reconcile two paradigms of a medical model that espouses a diagnosis-label-intervention axis versus a more ecological model that reverts back to perceived emphasis on the environment and by extension, the parents' influence or parenting in the child's environment (Gutkin, 2012). Future research might consider building on research that looks at the types of interventions or support parents most desire for their children (Kasilingam et al., 2019) and ask if this support could be provided without a diagnosis, it would be sufficient or even, more desirable than following a diagnosis-label paradigm.

Another consideration for future research with parents is to investigate what features of their child's neurodivergence have the greatest impact on their family’s functioning and quality of life. Research suggests that challenging behaviour may more difficult for families than the actual diagnostic characteristics of autism or ADHD (Ooi et al., 2016). It might be that the diagnostic criteria makes up the more positive aspects of reported family daily life, such as a specific interest (autism), and that other behaviours not indicated by diagnosis are more challenging for family daily-life. A meta synthesis on parenting an autistic child suggests this to be the case (Ooi et al., 2016). Therefore, it might be in case complexity or the additional co-occurring challenges like sleep and behaviour require greater attention to support families and children in education.

Conclusion

Autism and ADHD have significant co-occurrence rates. This population has been understudied due to previous restrictions on a dual diagnosis specified in diagnostic manuals. Preliminary research has shown that the interaction of these two conditions produces unique effects on diagnosis, interventions and outcomes for these children. The particular focus of this research has been the effect of autism + ADHD on diagnosis.

The results of the studies presented in this thesis demonstrate a multifaceted approach in attempting to understand the phenomena of the diagnosis of co-occurring autism + ADHD. It is likely that the challenges around diagnosis for autism + ADHD are compounded by developmental differences, societal difficulties of the diagnosis system, and the professional difficulties of diagnosing complex neurodevelopmental cases. A differing developmental pattern, reduced diagnostic-triggering concerns, and later specialist seeking has been shown to affect this co-occurring group. In addition, like other parents' experiences of the diagnosis of autism and ADHD, the perceived adversarial system to diagnosis creates delays and added insecurities to identity. Finally, from a clinician's point of view, an approach to a complex neurodevelopmental case requires both specific and more comprehensive assessments suggesting a time sensitive and heavily resourced process. The opportunities for immediate support are mostly referrals into other systems, as corroborated by the thematic analysis of parents' reported experience.

The identification of a number of possible causes to a delay in autism diagnosis when ADHD is present could allow for change to occur, and, in the longer term the reduction, or even eradication of the delay. The study of co-occurrence, and co-occurrence of autism + ADHD in particular, remains in its infancy, and a further development of findings and research is needed in this area.

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APPENDIX B

PARTICIPANT INFORMATION AND CONSENT FOR STUDY 1 AND 2

Welcome to the research study -

Parents of children who have autism and/or ADHD diagnoses

INFORMATION FOR PARTICIPANTS

You are invited to take part in this research. Please read this information before deciding whether or not to take part. If you decide to participate, thank you. If you decide not to participate, thank you for considering this request. We are interested in understanding what it is like to have a child diagnosed with either autism, attention deficit hyperactivity disorder (ADHD) or both diagnoses.

Who am I?

My name is Willow Sainsbury and I am a Doctoral student at Victoria University of Wellington.

What is the aim of the project?

This project aims to understand what it is like to get a diagnosis or multiple diagnoses for your child in Aotearoa. It also aims to understand the timing and type of your first concern you had for child and if that was related to his/her/they later diagnosis. This research has been approved by the Victoria University of Wellington Human Ethics Committee *[*Approval number 28993*]*.

How can you help?

You can participate if your child is 18 years of age or younger and has received a diagnosis of attention deficit hyperactivity disorder (ADHD) or autism spectrum disorder (ASD) or both.

If you agree to take part, you will complete a short survey.

The survey will take around 15 minutes to complete. This survey is in three parts:

- Part 1 asks three questions about background information so we know we have parents of different backgrounds answering.
- Part 2 asks about when you first had concerns about your child's development and the age of diagnosis for your child.
- Part 3 asks about experiences of getting a diagnosis.

What will happen to the information you give?

This research is anonymous. This means that nobody, including the researchers will be aware of your identity. By answering it, you are giving consent for us to use your responses in this research. Your answers will remain completely anonymous and unidentifiable. Once you submit the survey, it will be impossible to retract your answer. Please do not include any personal identifiable information in your responses.

Data will be kept until the end of 2023.

A summary of research findings can be accessed directly by emailing:

willow.sainsbury@vuw.ac.nz

Key research findings will also be shared with key organisations involved with diagnosis. Other proposed publications include academic or professional journals, conferences and a thesis in the Victoria University Library.

If you have any questions or problems, who can you contact?

If you have any questions, either now or in the future, please feel free to contact either my supervisor or me:

Student:	**Supervisor:**
Name: Willow Sainsbury	Name: Hannah Waddington
University email address:	Role: Lecturer in Education (Psychology)
willow.sainsbury@vuw.ac.nz	*hannah.waddington*@vuw.ac.nz

Human Ethics Committee information

If you have any concerns about the ethical conduct of the research you may contact the Victoria University of Wellington HEC Convenor: Associate Professor Judith Loveridge. Email hec@vuw.ac.nz or telephone +64-4-463 6028.

By clicking the button below, you acknowledge:

- You are 18 years of age or over.
- You are the primary carer of a child with a diagnosis of ASD and/or ADHD
- You and your child are currently living in New Zealand.

- I consent, begin the study
- I do not consent, I do not wish to participate

APPENDIX C

PARTICIPANT INFORMATION AND CONSENT FOR STUDY 3

Welcome to the research study - Clinicians (paediatricians, psychologists, psychiatrists) Diagnosing 4-and-8-year olds

INFORMATION FOR PARTICIPANTS

You are invited to take part in this research. Please read this information before deciding whether or not to take part. If you decide to participate, thank you. If you decide not to participate, thank you for considering this request. We are interested in understanding concerns and pathways you might consider when evaluating complex behaviour presentations in children.

Who am I?

My name is Willow Sainsbury and I am a Doctoral student at Victoria University of Wellington. This research project is work towards my thesis.

What is the aim of the project?

Using two vignettes, this project aims to understand what key features might be of concern to a diagnostician of neurodevelopmental conditions and what, if any, would be your possible next lines of investigation.

Your participation will support research by helping referrals to be better written with more relevant information for a possibly faster triage and improved diagnosis pathways. This

research has been approved by the Victoria University of Wellington Human Ethics Committee [Approval number 28993].

How can you help?

You have been invited to participate because you are involved with diagnosing children with learning or developmental diagnoses. If you agree to take part, you will complete five questions based on brief hypothetical descriptions (vignettes) of a child aged 4 and 8-years-old.

It is difficult to generalise from a vignette, however the purpose of this study is to get a general idea of what Clinicians might consider as next steps for this hypothetical child.

The survey is divided into two parts.

Part 1: A few details about your practice to ensure we have a range of Clinicians.

Part 2: Hypothetical vignettes and five short questions

The study should take you around **15 minutes to complete**.

What will happen to the information you give?

This research is anonymous. This means that nobody, including the researchers will be aware of your identity. By answering it, you are giving consent for us to use your responses in this research. Your answers will remain completely anonymous and unidentifiable. Once you submit the survey, it will be impossible to retract your answer. Please do not include any personal identifiable information in your responses.

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A summary of research findings can be accessed directly by emailing:

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Key research findings will also be shared with key organisations involved with diagnosis. Other proposed publications include academic or professional journals, conferences and a thesis in the Victoria University Library.

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If you have any questions, either now or in the future, please feel free to contact either my supervisor or me:

Student:	**Supervisor:**
Name: Willow Sainsbury	Name: Hannah Waddington
University email address:	Role: Lecturer
willow.sainsbury@vuw.ac.nz	School: Education (Psychology)
	hannah.waddington@vuw.ac.nz

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If you have any concerns about the ethical conduct of the research you may contact the Victoria University of Wellington HEC Convenor: Associate Professor Judith Loveridge. Email hec@vuw.ac.nz or telephone +64-4-463 6028.

By clicking the button below, you acknowledge:

- Your participation in the study is voluntary.
- You are 18 years of age or over.
- You are currently practicing in New Zealand.
- You are aware that you may choose to terminate your participation of this survey at any time before submitting it.

- I consent, begin the study
- I do not consent, I do not wish to participate

APPENDIX D

PARENT SURVEY

Welcome to the research study -

Parents of children who have autism and/or ADHD diagnoses

You are invited to take part in this research. Please read this information before deciding whether or not to take part. If you decide to participate, thank you. If you decide not to participate, thank you for considering this request. We are interested in understanding what it is like to have a child diagnosed with either autism, attention deficit hyperactivity disorder (ADHD) or both diagnoses.

Who am I?

My name is Willow Sainsbury and I am a Doctoral student at Victoria University of Wellington.

What is the aim of the project?

This project aims to understand what it is like to get a diagnosis or multiple diagnoses for your child in Aotearoa. It also aims to understand the timing and type of your first concern you had for child and if that was related to his/her/they later diagnosis. Your answers might help with improving the diagnosis pathway. This research has been approved by the Victoria University of Wellington Human Ethics Committee [Approval number 28993].

How can you help?

You can participate if your child is 18 years of age or younger and has received a diagnosis of

attention deficit hyperactivity disorder (ADHD) or autism spectrum disorder (ASD) or both.

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The survey will take around 15 minutes to complete. This survery is in three parts:

- Part 1 asks three questions about background information.
- Part 2 asks about when you first had concerns about your child's development and the age of diagnosis for your child.
- Part 3 asks about experiences of getting a diagnosis.

What will happen to the information you give?

This research is anonymous. This means that nobody, including the researchers will be aware of your identity. By answering it, you are giving consent for us to use your responses in this research. Your answers will remain completely anonymous and unidentifiable.

You may choose to terminate your participation of this survey at any time before submitting it, if you do so your answers will not be become part of the research.

If you have any questions or problems, who can you contact?

If you have any questions, either now or in the future, please feel free to contact either my supervisor or me:

Student:

Name: Willow Sainsbury

University email address: willow.sainsbury@vuw.ac.nz

Supervisor:

Name: Hannah Waddington

Role: Lecturer

School: Education (Psychology)

Phone:

hannah.waddington@vuw.ac.nz

Human Ethics Committee information

If you have any concerns about the ethical conduct of the research you may contact the Victoria University of Wellington HEC Convenor: Associate Professor Judith Loveridge. Email hec@vuw.ac.nz or telephone +64-4-463 6028.

By clicking the button below, you acknowledge:

- You are 18 years of age or over.
- You are the primary carer of a child with a diagnosis of ASD and/or ADHD
- You and your child are currently living in New Zealand.

○ I consent, begin the study (1)

○ I do not consent, I do not wish to participate (2)

End of Block: Informed Consent

Start of Block: Demographics Base/Universal

Q 1 What is your relationship to the child with a diagnosis?

- ○ Mother (1)
- ○ Father (2)
- ○ Caregiver (3)
- ○ Legal guardian (4)

Q 2 What is the gender of your child?

- ○ Male (1)
- ○ Female (2)
- ○ Non-binary (3)
- ○ Prefer not to say (4)

Q 3 How old is your child?

_____ Year (1)

_____ Months (2)

Q4 Does your child have any brothers and sisters (siblings)?

- ☐ No other siblings (1)
- ☐ Sibling(s) of the same age (e.g. a twin) (2)
- ☐ Older sibling(s) (3)
- ☐ Younger sibling(s) (4)

☐ Prefer not to say (5)

Q5 Which ethnic group do you belong to:

☐ Māori (1)

☐ NZ European (2)

☐ Asian (3)

☐ Cook Island Māori (4)

☐ Tongan (5)

☐ Samoan (6)

☐ Niuean (7)

☐ Prefer not to say (8)

☐ Other (9) _____

Q6 What was your approximate total household income last tax year?

- ○ Less than $10,000 (1)
- ○ $10,000 to $19,999 (2)
- ○ $20,000 to $29,999 (3)
- ○ $30,000 to $39,999 (4)
- ○ $40,000 to $49,999 (5)
- ○ $50,000 to $59,999 (6)
- ○ $60,000 to $69,999 (7)
- ○ $70,000 to $79,999 (8)
- ○ $80,000 to $89,999 (9)
- ○ $90,000 to $99,999 (10)
- ○ $100,000 to $149,999 (11)
- ○ $150,000 or more (12)
- ○ Prefer not to say (13)

Q7 What is the highest level of school you have completed or the highest degree you have received?

- ○ Attended school (1)
- ○ High school graduate (2)
- ○ University degree (3)
- ○ Master's degree (4)
- ○ Doctoral degree (5)
- ○ Prefer not to say (6)
- ○ Other (7) _____

Q8 Do you recall anything unusual about your child's development or behaviour during the first six months of his/her life?

- ○ Yes (1)
- ○ No (2)

Q9 Now, think of the period between 6 and 12 months, can you recall anything unusual about your child's development or behaviour during that time?

- ○ Yes (1)
- ○ No (2)

Please select all aspects of unusual development that applied to your child	**Birth - 6 months of age Tick all that apply**	**7 - 12 months of age Tick all that apply**
Delayed speech or vocalisations	☐	☐
No speech or vocalisations	☐	☐
Poor language understanding	☐	☐
Loss of language	☐	☐
Delayed social communication	☐	☐
No social communication	☐	☐
Unusual eye contact	☐	☐
Poor social interaction	☐	☐

Lack of response to people	☐	☐
Repetitive movements	☐	☐
Need for routines/rituals	☐	☐
Restricted interests	☐	☐
Obsessed with parts of an object	☐	☐
Lack of attention	☐	☐
Hyperactivity	☐	☐
Inaction (passivity)	☐	☐
Very sensitive to sounds, touch etc.	☐	☐

Under sensitive to sound, touch etc.	☐	☐
Delay in movement (motor delay - fine and gross skills)	☐	☐
Decreased muscle tone	☐	☐
Increased muscle tone	☐	☐
Swallowing and sucking difficulties	☐	☐
Difficulties with eating	☐	☐
Difficulties with sleeping	☐	☐
Tantrums/ opposition	☐	☐
Difficult to soothe	☐	☐

Aggression/violence	☐	☐
Self-harm	☐	☐
Extreme attachment to caregiver	☐	☐
Another disorder	☐	☐
Sickness	☐	☐
Other	☐	☐

Skip To: Q11 If Please select all aspects of unusual development that applied to your child at birth to 6 months... : Birth - 6 months of age = Other [Tick all that apply]

Q11 If there were any other unusual aspects of your child's development, please describe below.

☐ From birth to 6 months (1)

_____

☐ 6 -12 months of age (2)

_____

Q12 How old was your child when you first had concerns about your child's development and you needed to see a specialist?

(If you cannot remember clearly, please give an estimate)

_____ Year (1)

_____ Months (2)

Q13 What were the concerns that led you to seek specialist help?

_____

_____

_____

Q14 How many specialists/professionals did you see prior to getting a diagnosis? (tick all that apply).

- ☐ GP (1)
- ☐ Ministry of education professional (2)
- ☐ Speech & language therapist (3)
- ☐ Psychologist (4)
- ☐ Psychiatrist (5)
- ☐ Paediatrician (6)
- ☐ Other, please explain (7)

_____

Q15 Which of the following diagnoses have been given to your child?

- ○ Autism and ADHD (1)
- ○ Autism (2)
- ○ ADHD (3)

Skip To: Q17 If Which of the following diagnoses have been given to your child? = Autism

Skip To: Q16 If Which of the following diagnoses have been given to your child? = ADHD

Skip To: Q16 - 17 If Which of the following diagnoses have been given to your child? = Autism and ADHD

Q16 How old was your child when the ADHD diagnosis was given?

(If you cannot remember clearly, please give an estimate)

_____ Year (1)

_____ Months (2)

Q17 How old was your child when the autism diagnosis was given?

(If you cannot remember clearly, please give an estimate)

_____ Year (1)

_____ Months (2)

Q18 In your opinion, which diagnosis best explains the behaviours of your child?

- ○ Autism symptoms (social communication difficulties, fixed rigid behaviour) (1)
- ○ ADHD symptoms (Inattentive, and/or hyperactive, and/or impulsive) (2)
- ○ Both (3)
- ○ Neither (4)

Skip To: Q19 If In your opinion, which diagnosis best explains the behaviours of your child? = Neither

Q19 In your opinion which diagnosis or explanation best describes the behaviours of your child?

_____

_____

_____

_____

_____

Q20 Does your child have any other diagnoses?

- ☐ Sensory processing disorder (1)
- ☐ Epilepsy/seizures (2)
- ☐ Oppositional defiant disorder (3)
- ☐ Intellectual disability/global delay (4)
- ☐ Learning disability/dyslexia (5)
- ☐ Other (6) _____

Q21 Has your child ever received any previous diagnoses that now no longer apply? If yes, please list the diagnosis he/she was given, that now no longer applies.

- ○ Yes (1) _____
- ○ No (2)

Q22 What behaviours of your child provide the most challenges for your family/child's life?

_____

_____

_____

_____

_____

Q23 What was helpful during the diagnostic process?

_____

_____

_____

_____

_____

Q24 What was unhelpful about the diagnostic process?

_____

_____

_____

_____

_____

Q25 What else would you like researchers to know about your experience of getting a diagnosis for your child?

_____

_____

_____

_____

If you consent to submitting your responses, please press the right hand arrow.

You will not be able to review your answers once you push the arrow.

End of Block: Block 2

APPENDIX E

CLINICIAN SURVEY

Welcome to the research study -

Clinicians (paediatricians, psychologists, psychiatrists) Diagnosing 4-and-8-year olds

You are invited to take part in this research. Please read this information before deciding whether or not to take part. If you decide to participate, thank you. If you decide not to participate, thank you for considering this request.

Who am I?

My name is Willow Sainsbury and I am a Doctoral student at Victoria University of Wellington. This research project is work towards my thesis.

What is the aim of the project?

Using two vignettes, this project aims to understand what key features might be of concern to a clinician and what, if any, would be possible next lines of investigation.

We are interested in understanding concerns and pathways you might consider when evaluating complex behaviour presentations in children. This research has been approved by the Victoria University of Wellington Human Ethics Committee [Approval number 28993].

How can you help?

You have been invited to participate because you are involved with diagnosing children with learning or developmental diagnoses. If you agree to take part, you will complete five questions based on brief hypothetical descriptions (vignettes) of a child aged 4 and 8-years-old.

It is difficult to generalise from a vignette, however the purpose of this study is to get a general idea of what Clinicians might consider as next steps for this hypothetical child.

The survey is divided into two parts.

Part 1: A few details about your practice to ensure we have a range of Clinicians.

Part 2: Hypothetical vignettes and five short questions

The study should take you around **15 minutes to complete**.

What will happen to the information you give?

This research is anonymous. This means that nobody, including the researchers will be aware of your identity. By answering it, you are giving consent for us to use your responses in this research. Your answers will remain completely anonymous and unidentifiable. Once you submit the survey, it will be impossible to retract your answer. Please do not include any personal identifiable information in your responses. You can skip any questions you do not wish to answer.

If you have any questions or problems, who can you contact?

If you have any questions, either now or in the future, please feel free to contact either my supervisor or me:

Student:

Name: Willow Sainsbury

University email address:

willow.sainsbury@vuw.ac.nz

Supervisor:

Name: Hannah Waddington

Role: Lecturer

School: Education (Psychology)

hannah.waddington@vuw.ac.nz

Human Ethics Committee information

If you have any concerns about the ethical conduct of the research you may contact the Victoria University of Wellington HEC Convenor: Associate Professor Judith Loveridge. Email hec@vuw.ac.nz or telephone +64-4-463 6028.

By clicking the button below, you acknowledge:

Your participation in the study is voluntary.

You are currently practicing in New Zealand

You are aware that you may choose to terminate your participation of this survey at any time before submitting it.

- ○ I consent, begin the study (1)
- ○ I do not consent, I do not wish to participate (2)

End of Block: Default Question Block

Start of Block: Block 1

Q28 Do you or have you diagnosed children in New Zealand?

- ○ Yes (1)
- ○ No (2)

Q5 Which ethnic group do you belong to:

- ☐ Māori (1)
- ☐ NZ European (2)
- ☐ Asian (3)
- ☐ Cook Island Māori (4)
- ☐ Tongan (5)
- ☐ Samoan (6)
- ☐ Niuean (7)
- ☐ Other (8) _____
- ☐ Prefer not to say (9)

Q6 What is your role?

- ○ Paediatrician (1)
- ○ Psychologist (2)
- ○ Psychiatrist (3)
- ○ Prefer not to say (4)
- ○ Other (5) _____

Q7 How long have you been practicing?

- ○ 1-2 years (1)
- ○ 3 -4 years (2)
- ○ 5-9 years (3)
- ○ 10+ years (4)
- ○ Prefer not to say (5)

Q30 Did you do most of your training in New Zealand?

- ○ Yes (1)
- ○ No (2)
- ○ Prefer not to say (3)

Q8 Which sector do you work in?

- ○ Public (1)
- ○ Private (2)
- ○ Both (3)
- ○ Prefer not to say (4)

Q9 Within your scope of practice, do you have a particular specialty interest?

- ○ Yes, Please describe your interest (1)

_____

- ○ No (2)
- ○ Prefer not to say (3)

End of Block: Block 1

Start of Block: Block 2

Q10 **Vignette 1. Brief history in the referral:** Jack is a four-year-old boy. He has recently had his hearing and eyesight checked with no concerns. He had delayed speech and he has had a short course of speech and language therapy (SLT). He is now speaking in three word phrases. He can name all his colours, count to 5, but jumbles his numbers after 5. His parents separated when he was 3 years old. The early childhood centre states that he does not participate well with the rest of the children, particularly during mat time. He will latch on to one adult or child for the day and direct them by lifting their hand to what he wants. He likes the dinosaurs and he will place them in different parts of the centre and he sometimes becomes very upset if the dinosaurs are then moved, which can lead to him throwing things. He will often try to escape the childhood centre, seeing it as a game by taking any moment to run through the gate and run off down the driveway. He likes roaring like a dinosaur and hiding under tables.

Q11 In Jack's referral, what would you consider to be a concern?

	Important concern (1)	A concern (2)	Moderate concern (3)	Slight concern (4)	Not at all important (5)
Language delay (1)	○	○	○	○	○
Cognitive knowledge (2)	○	○	○	○	○
Parent separation (3)	○	○	○	○	○
Lack of participation (4)	○	○	○	○	○
Person as tool (5)	○	○	○	○	○
Placing objects (6)	○	○	○	○	○
Becomes very upset (7)	○	○	○	○	○
Elopement (8)	○	○	○	○	○

Loud vocalisations (9)	○	○	○	○	○
Hiding (10)	○	○	○	○	○

Q15 What assessment (s) would you consider for this child?

○ No assessment (1)

○ Assessments - please name the assessment you might consider? (2)

_____

Q13 What age might you recommend the assessment(s) be done?

- ○ Age 4 (immediately) (1)
- ○ Age 5 (when he starts school) (2)
- ○ Age 6 (3)
- ○ Age 7-10 (4)
- ○ In high school (5)
- ○ If different assessments at different time points, please explain (6)

_____

Q14 Without more information, what diagnosis/diagnoses might you be considering for this child?

_____

_____

_____

_____

_____

Q16 Would you consider recommending any services/interventions immediately?

- ○ Yes, if so what services or intervention would you recommend? (1)

  _____

- ○ No (2)

End of Block: Block 2

Start of Block: Block 3

Q24 **Vignette 2. Brief history in the referral:** Thomas is an 8 year old boy. He has difficulty sleeping at night, and his mother reports poor sleep from an early age. He had his hearing, eyesight checked and he had a cognitive assessment which raised no concerns with a reported IQ score in the average range. He likes reading his favourite books and his language appears at his age level. He has trouble making friends at school, and he has started a social skills program with his teacher and teacher aide in a small group. The teacher reports that he is often forgets what task he should be working on and instead often shouts a swear word to be funny for his classmates. He has trouble following classroom instructions. He does not appear interested in much at school, but at home his mother reports that he loves playing lego. He gets told off frequently for his voice which is either shouting or a whisper. The other children typically avoid Thomas, but he tends to move from his seat to get closer to his classmates to see what they are doing.

Q18 In Thomas' referral, what would you consider to be a concern?

	Important concern (1)	A concern (2)	Moderate concern (3)	Slight concern (4)	Not at all important (5)
Difficulty sleeping (1)	○	○	○	○	○
Average IQ (2)	○	○	○	○	○
Difficulty making friends (3)	○	○	○	○	○
Forgets what to to do (4)	○	○	○	○	○
Swearing (5)	○	○	○	○	○
Difficulty following classroom instructions (6)	○	○	○	○	○
Low interest (7)	○	○	○	○	○
Shouting/whispering (8)	○	○	○	○	○
Not socially accepted (9)	○	○	○	○	○

Leaves seat (10)	○	○	○	○	○

Q20 What assessment (s) would you consider for this child?

- ○ No assessment (1)
- ○ Assessments - please name the assessment you might consider? (2)

_____

Q21 What age might you recommend the assessment(s) be done?

- ○ Age 8 (immediately) (1)
- ○ Age 9 -10 (2)
- ○ Age 11 -13 (intermediate school) (3)
- ○ In high school (4)
- ○ If different assessments at different time points, please explain (5)

_____

Q22 Without more information, what diagnosis/diagnoses might you be considering for this child?

_____

_____

_____

_____

_____

Q23 Would you consider recommending any services/interventions?

☐ Yes, if so what services or intervention would you recommend? (1)

_____

☐ No (2)

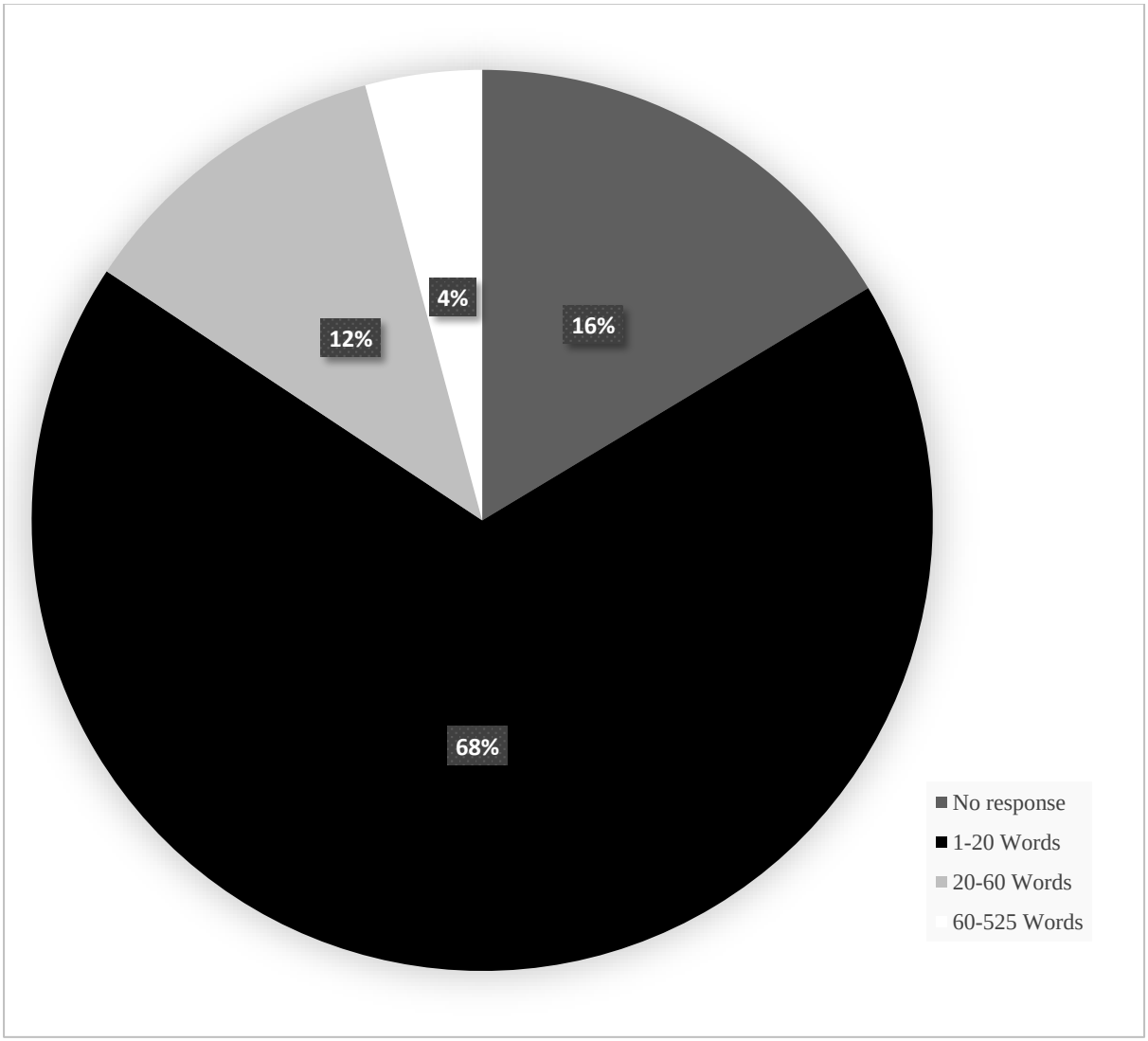
Q26 If you consent to submitting your responses, please press the right hand arrow.

You will not be able to review your answers once you push the arrow.

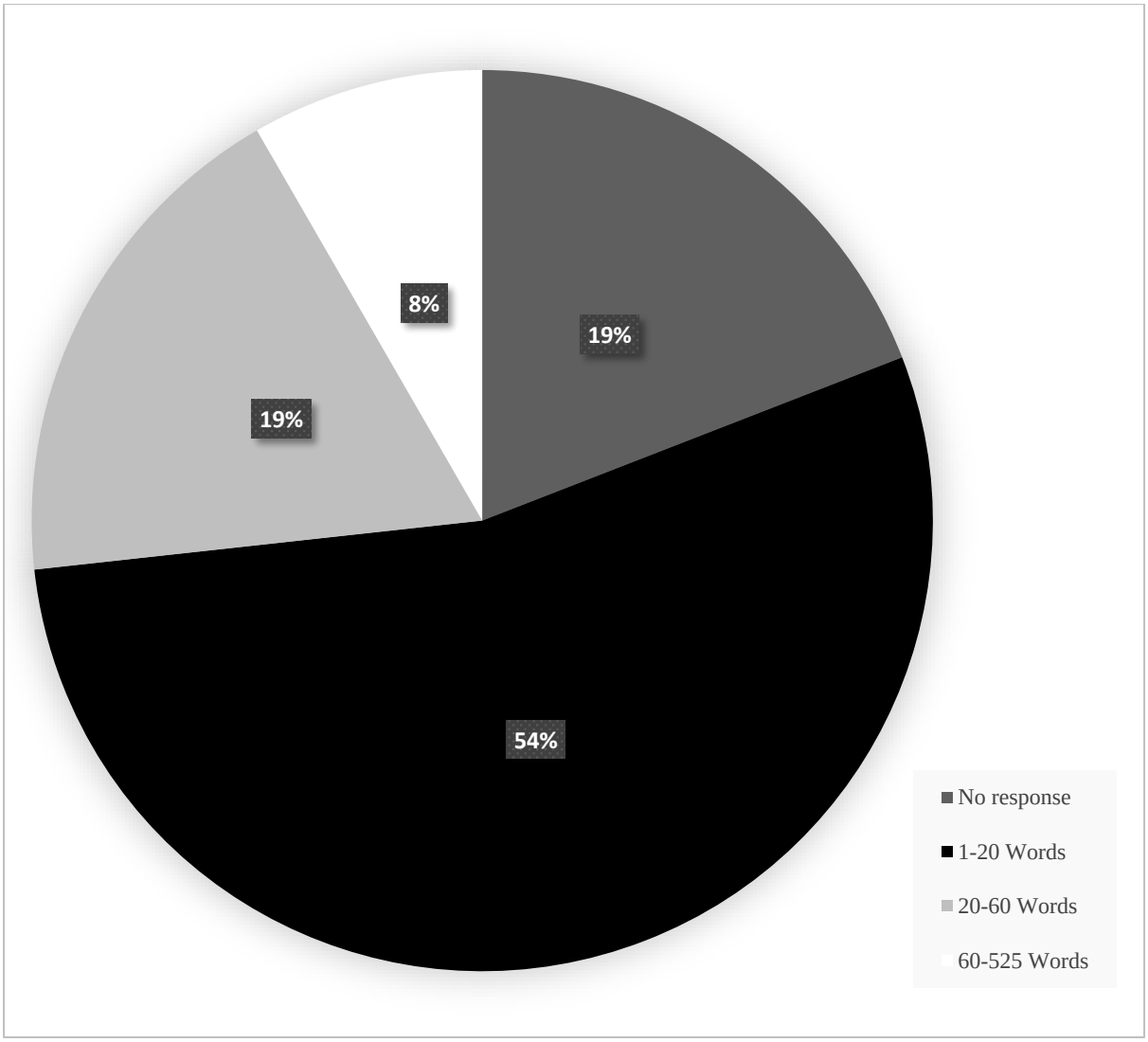
End of Block: Block 3

APPENDIX F

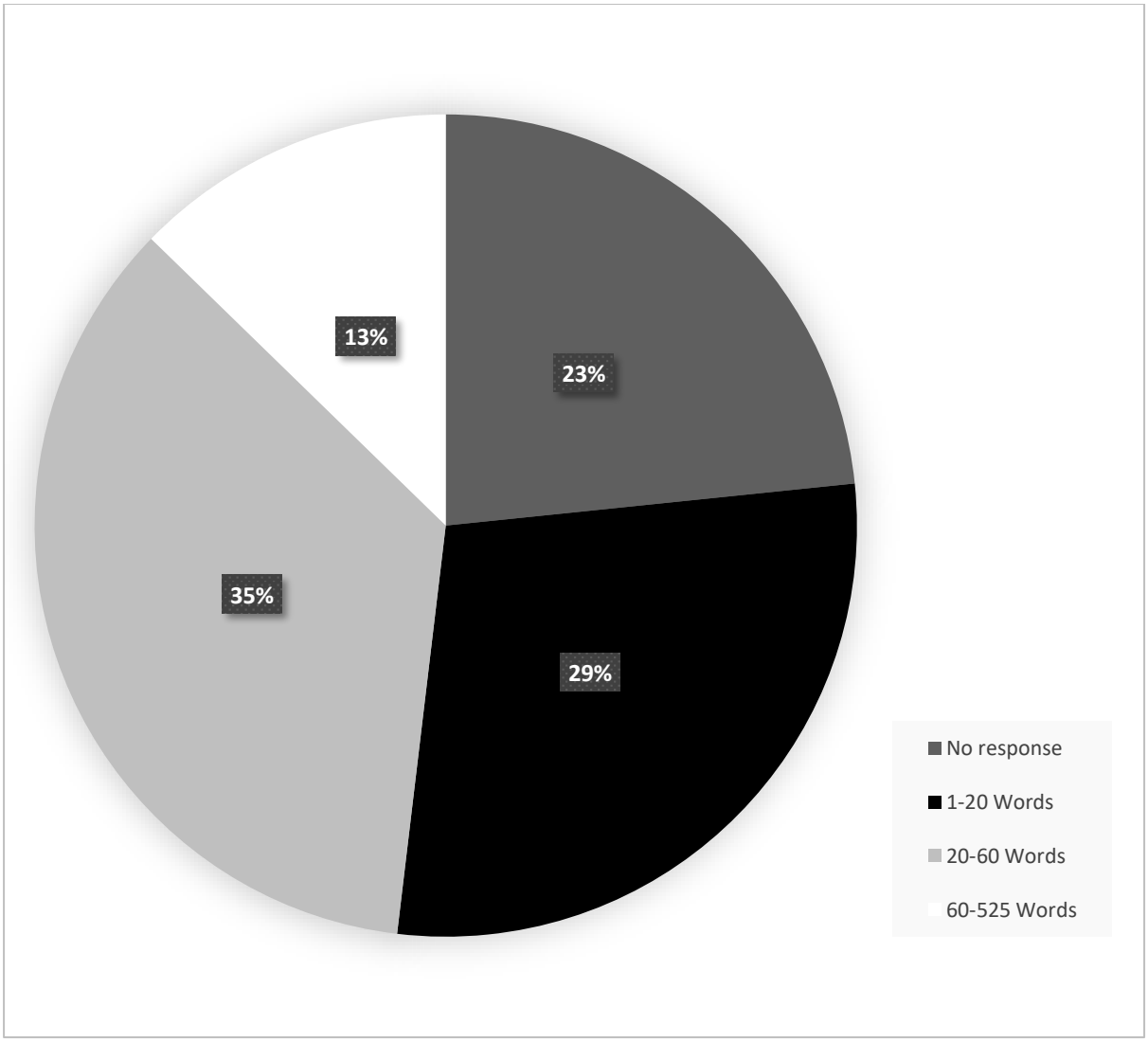
SUPPLEMENTARY MATERIAL FOR STUDY 2



Supplementary Figure 1. Response rate to the question: What was helpful during the diagnosis process?



Supplementary Figure 2. Response rate to the question: What was unhelpful during the diagnosis process?



Supplementary Figure 3. Response rate to the question: What would you like researchers to know about the diagnosis process.

APPENDIX G

RESEARCH SNAPSHOT FOR PARTICIPANTS

Research Snapshot - AUTISM, ADHD and AUTISM+ADHD

AIM

To understand what might look different between children with autism, ADHD, autism+ADHD in the experience of diagnosis.

BACKGROUND

Autistic+ADHD children have a delayed autism diagnosis compared to autistic children.

Autistic+ADHD children have an early ADHD diagnosis compared to ADHD children.

WHO TOOK PART?

288 PARENTS PARTICIPATED.

- 111 had an autistic child
- 93 had an ADHD child
- 84 had an autistic+ADHD child

Thank you to all those parents, who participated. ☺

HOW WE DID THE RESEARCH?

We asked parents of children with autism and/or ADHD children under the age of 18, to answer questions about:

- Themselves and their child
- The timing and development of concerns
- What was helpful/unhelpful

WHAT WE FOUND

Differences in age of diagnosis:

-Autism diagnosis was 3.2 years later if the child had co-occurring ADHD.
-ADHD diagnosis was 1.4 years earlier if the child also had co-occurring autism.

Differences in number and type of development concerns during a child's first year:

-No significant number of parent-reported early development concerns between the autism and autism + ADHD group, but significantly less in for the ADHD group.

-The autism group reported more language and social characteristics than the autism + ADHD group.

-All three groups reported similar amounts of behaviour/temperament, sleeping and feeding concerns.

Differences in the diagnostic pathway:

-The age a clinician was sought was earlier for the autism group, but similar for the ADHD and autism + ADHD group.

-The majority of the autism group reported an autism characteristic to a clinician while the majority of the ADHD and autism + ADHD group reported presenting to a clinician with a characteristic that did not fit with autism or ADHD.

-This pattern also fit with a longer time to diagnosis if the presenting characteristic did not fit a diagnosis criterion.

-The autism + ADHD group saw more types of specialist and waited longer than the autism group

Parents' experience of diagnosis for their child:

-Parents' reported experiencing two battles; one for diagnosis and the other for support for their children.

-Application of criteria and thresholds for diagnosis or support were often frustrating and defeatist.

-Parenting courses acted as 'gate keepers' to diagnosis, attributed blame to parents, and were often unhelpful.

-The autism group reported that a celebration of autism as neurodivergent was beneficial.

-The ADHD group reported being told their child was "too young" for diagnosis and medication was often given in isolation.

WHO DID THIS RESEARCH AND WHERE CAN YOU FIND OUT MORE?

Willow Sainsbury: willow.sainsbury@vuw.ac.nz

Sainsbury, W.J., Carrasco, K., Whitehouse, A.J.O., Waddington, H. (2022) Parent-reported Early Atypical Development and Age of Diagnosis for Children with Co-occurring Autism and ADHD. *Journal of Autism and Developmental Disorders,* 1-12. https://doi.org/10.1007/s10803-022-05488-0

Sainsbury, W.J., Carrasco, K., Whitehouse, A.J.O., Waddington, H. (2022) Parent experiences of diagnosis for children with autism, ADHD, or both conditions. *International Journal of Developmental Disabilities,* (In Press).

VICTORIA UNIVERSITY OF WELLINGTON TE HERENGA WAKA