An Exploration of the Presence and Behavioural Manifestation of Anxiety in Children and Young Adults with Autism Spectrum Disorder and Intellectual Disability

Author

Appleton, Helen D

Published

2022-11-21

Thesis Type

Thesis (PhD Doctorate)

School

School Educ & Professional St

DOI

10.25904/1912/4710

An Exploration of the Presence and Behavioural Manifestation of Anxiety in Children and Young Adults with Autism Spectrum Disorder and Intellectual Disability

Helen Dawne Appleton

BSc, BA, GDS(Psych)

Autism Centre of Excellence

School of Education and Professional Studies

Griffith University

Submitted in fulfillment of the requirements of the degree of

Doctor of Philosophy

July 2022

Abstract

Individuals with autism spectrum disorder (ASD) display a pattern of social communication deficits, restricted and repetitive behaviours, and information processing difficulties that leave them particularly vulnerable to developing anxiety. The process of diagnosing anxiety is confounded by the very nature of the communication difficulties found in this population and is further exacerbated when a co-occurring intellectual disability (ID) is also present. As part of this research, a scoping review was conducted into the current research available directly addressing how anxiety is identified or diagnosed in individuals with ASD and ID. This review found very limited research relevant to this population and identified the lack of specifically standardised anxiety measurement tools as a particular issue, as well as the need to consider different symptom criteria in order to more accurately diagnose anxiety in this unique population, with behavioural markers being a useful consideration. For this study, the dynamic model of autism (Bartak et al., 2006) was selected as a theoretical framework that explains how anxiety develops in individuals with ASD without excluding those with a co-occurring ID. Many other theories of anxiety development in autism neglect those with ASD and ID. In this research two key concepts are considered: firstly, the identification and testing of this theoretical framework for understanding how anxiety is related to ASD and ID, and secondly, investigation of behavioural profiles that could be used as markers to identify anxiety in individuals with ASD and ID. Through four hierarchical multiple regressions, the dynamic model of autism was found to offer an explanation for how anxiety develops in individuals with ASD and ID. This theory proposes that it is the very core characteristics of ASD that result in an individual's vulnerability to developing anxiety, and it may be that those with co-occurring ID are even more at risk given their lack of ability to develop alternative strategies. Bayesian model averaging was used to investigate behavioural patterns predicting anxiety in individuals with ASD and ID. Within this study, several behavioural markers were found to be predictive of increased anxiety, including increased insistence on sameness, increased sensory sensitivity, and increased problem behaviour. By considering that anxiety may be the potential cause of problem behaviours we may in turn have more success with addressing ASD behavioural escalations. Increased recognition of the mental health experiences and the need for appropriate treatment and support for this under-researched population could lead to better quality of life outcomes in general and in the longer term.

Statement of Originality

This work has not previously been submitted for a degree or diploma in any university. To the best of my knowledge and belief, the thesis contains no material previously published or written by another person except where due reference is made in the thesis itself.

(Signed) [redacted] (Date)__21/7/22__________

Helen Appleton

Table of Contents

List of Tables

List of Figures

Acknowledgements

This PhD has been, at times, the most significant part of my life for over 8 years, representing a considerable academic, professional, and personal challenge. I have learnt more about autism, my work, and my own abilities than I thought possible through this process. It has been exhausting and frustrating to complete such arduous research alongside the full-time demands of working within an organisation like Giant Steps. Yet without the day-to-day reminders of why the research was so dearly needed, my tenacity alone may not have been enough to complete it.

To all the ***students, clients, and families at Giant Steps Australia***, I express my heartfelt thanks for their participation in this research, for their ongoing support of my work, for the many inquiries and offers of meals and help over the years. That you could offer me support despite your own daily battles and troubles has always been humbling. It is an honour to be part of your children's lives and to see who they become as they grow. I hope this research is part of a larger movement towards understanding of our unique population and offering them better opportunities in the future.

To ***all the staff at Giant Steps Australia,*** past and present—thank you for your time, patience, and many hours filling out questionnaires. Thank you for listening to me as I worked through my thoughts and theories and found a way to bring them to life. To the ***Giant Steps Board, executive*** and ***leadership teams***, who over the last 8 years has offered me a huge amount of support and encouragement, allowing me time away and filling in when needed. Thanks especially to ***Kerrie Nelson*** and ***Andrew Frakes*** who have offered unquestioning support, offering me time even when I was terrible at taking it.

I am immeasurably grateful to my supervisory team, who have battled through with me for so long. To ***Greta Ridley***, ***Alison Waters*** and ***Trevor Clark*** who were part of the early discussions, thank you for your input and guidance. I had a long-held dream to work with ***Prof. Jacqueline Roberts***—feeling seen and heard by a mentor you hold in such high esteem has been a remarkable experience. Thank you Jacqui for the many enjoyable conversations, the constructive feedback, and gentle prodding to stay ontrack; I appreciate it all more than you can know. Thank you of course to ***Dr Kate Simpson***, who has worked tirelessly to keep me on target and with an eye to the details that were not my forte. Thank you Kate for your patience, constructive feedback, and firm reminders of who I was writing for. Without you both, I truly would not have survived this process, nor have completed this research with anywhere near the sense of pride I currently do, so thank you so much for your tenacity in sticking with me.

Many thanks is needed for ***Clair Alston-Knox***, whose help with the Bayesian statistics in Chapter 6 was vital. Your ongoing help, availability, and support were remarkable, and access to Autostat was greatly appreciated.

Thank you to ***Elizabeth Stevens***, whose editing skills at the end of this process were professional, efficient and much needed. Thank you for your time and keen eye to detail.

While the process of completing a PhD is essentially a solo one, it would not be possible without the abundant support I have had from my closest friends and family. A most heartfelt thank you to my parents, ***Janice and Brenda Appleton***, whose ever-present support and belief in me is sometimes baffling. Your kindness, care, and constant interest, even from afar, is much appreciated and this thesis would have been impossible without it. To my dear big brother ***Darren***, whose own experience of ASD was my inspiration to venture into this world in the first place, thank you for the many conversations and utter belief in me. Watching your continual growth is a source of much pride.

Thank you to the many friends and colleagues who have supported, and simply given me time and understanding over the very long life of this thesis. Thank you to ***Erin Cowie***, who perhaps more than any other has heard my many updates and complaints through both work and travel together. Thank you for your patience and constant reassurance that all would end well.

To ***Dr Sarah McDonald,*** who is more sister than friend: Thank you will never be enough for the wisdom, insight, emotional support, and love you have given not only over the course of this thesis, but always. Thank you also for sharing your family, ***David***, ***Ruby,*** and ***Ted***, who have been my dinner and play breaks when they were sorely needed.

This PhD would not have occurred at all without ***Kerrie Nelson***. Without your blithe statement, "of course you should do a PhD H", I never would have contemplated it. Your complete faith in my ability gave me faith in myself. Thank you for the many years of being a sounding board, of questioning what needed to be questioned, and offering silence at just the right time. This PhD would not exist and I would not the person I am today without you.

ALL PAPERS INCLUDED ARE CO-AUTHORED

Acknowledgement of Papers Included in this Thesis

Included in this thesis is a paper in Chapter 3 which is co-authored with other researchers. My contribution to the co-authored paper is outlined at the front of the relevant chapter.

Chapter 3: I am the primary author on the published paper. The other two authors are my supervisors, who edited and offered feedback as to structure and format.

(Signed) ____[redacted]______ (Date)__21/7/22____________

Helen Appleton

(Countersigned) _________[redacted]_____ (Date)__21/7/22____________

Supervisor: Jacqueline Roberts

(Countersigned) [redacted] (Date)___21/7/22___________

Supervisor: Kate Simpson

Chapter 1: Introduction

Autism spectrum disorder (ASD) is a neurodevelopmental disorder, affecting an individual's social communication skills and leading to restricted and repetitive behaviours and interests (RRBs; American Psychiatric Association [APA], 2013). Intellectual disabilities (ID) are common among individuals with ASD and may exacerbate symptom severity and communication difficulties. The nature of autism and its associated processing difficulties leaves an individual vulnerable to comorbid psychiatric disorders, particularly anxiety, with reports that up to 84% of individuals with ASD experience clinically significant anxiety (Leyfer et al., 2006; Simonoff et al., 2008). Identifying and diagnosing anxiety in an individual with ASD and ID can be extremely difficult, as communication difficulties, symptom amplification, and diagnostic overshadowing can make differential diagnosis extremely problematic. The very definition of anxiety and the limitations of existing diagnostic tools may also impede a clear understanding of underlying psychiatric concerns for children with ASD and co-occurring ID. Without this understanding, programs are unlikely to adequately support the learning and engagement of an individual with ASD and ID, potentially resulting in social isolation, community exclusion, and increased burden of care for families.

Research Context

Anxiety is frequently associated with ASD, resulting in debate in recent research as to whether anxiety is a true comorbid disorder or a trait of ASD (Kerns & Kendall, 2013). The core characteristics of ASD—social communication deficits and RRBs—as well as the information processing characteristics, including weak central coherence, executive functioning deficits, and theory of mind difficulties, all seem to increase an individual's vulnerability to experiencing anxiety (Wood & Gadow, 2010). Differential diagnosis of anxiety and ASD is also hampered by the challenges inherent in differentiating the cognitive and behavioural markers of anxiety from those of ASD (Helverschou & Martinsen, 2011).

The presence of a co-occurring ID in individuals with ASD only increases the complexity inherent in identifying anxiety. Co-occurring ID has been found to amplify ASD symptom severity (Hill & Furniss, 2006), which is likely to result in further vulnerability to anxiety. However, specific research into the manifestation of anxiety for those with ASD suggests that those with ASD and no ID experience higher levels of anxiety symptoms than those with ASD and a co-occurring ID (Dickerson Mayes et al., 2011; Lecavalier, 2006). There is also the suggestion that the presence of ID with ASD results in a different pattern of manifestation of anxiety, as a result of the cognitive and communication difficulties experienced by this cohort (Sukhodolsky et al., 2008). Recent changes to the diagnostic criteria and process for ASD make it timely to consider how comorbidity of psychiatric conditions may be impacting on the individual, as a more holistic approach to diagnosis is supported by the *Diagnostic and Statistical Manual of Mental Disorders* (5th ed. [DSM-5]; APA, 2013). In previous editions (such as DSM-IV, APA, 1994), comorbid diagnosis with ASD was limited, but under DSM-5 the opportunities for comorbid diagnosis have been broadened to include attention deficit hyperactivity disorder, generalised anxiety disorder, separation anxiety, and social anxiety (APA, 2013).

It is unclear how the co-occurrence of ASD and ID affects the occurrence of comorbid anxiety and its manifestation in this population. It is also unclear if current definitions of and measurement tools for anxiety capture the presence of anxiety in individuals with ASD and ID.

Research Problem

Accurate diagnosis of anxiety in those with ASD and ID is hampered by a number of difficulties, including the very nature of the communication and information processing difficulties characteristic of ASD. Consideration should therefore be given to other possible methods of identifying and diagnosing anxiety, such as physiological changes (Helverschou & Martinsen, 2011) and behavioural clusters (Lecavalier, 2006). The combined characteristics of ASD and ID often result in additional stress and anxiety for the individual when faced with data collection of physiological markers of anxiety, therefore confounding the data collection if it can be collected at all (Romanczyk & Gillis, 2006); the focus, then, must turn to more observable behavioural patterns and phenotypes to offer insight into differential diagnosis of anxiety, ASD, and ID.

A number of theories explaining the relationship between ASD and anxiety have been offered. Most of these theories have a primarily cognitive basis, whereby the individual with ASD responds to and interprets the social world around them maladaptively, resulting in anxiety (Gotham et al., 2013; Wood & Gadow, 2010). However, the dynamic model of autism (Bartak et al., 2006) offers an explanation for the relationship between ASD and anxiety, which does not rely solely on the individual's cognitive ability to comprehend and interpret the social world around them. Instead, the dynamic model of autism postulates that it is the ongoing stressful interaction between an individual's core ASD characteristics and the environment, including interventions designed to remediate these ASD characteristics, that result in an anxiety disorder. Investigation of this model would offer insight into how anxiety develops in individuals with ASD and ID and the reasons they experience a particular vulnerability to this condition. The following research questions will be addressed:

1. Does the dynamic model of autism explain the relationship between anxiety and ASD for individuals with co-occurring ID?
2. Are there specific behavioural patterns for the expression of anxiety in individuals with ASD and ID?

Research Purpose and Significance

The purpose of this research is to consider comorbid anxiety in individuals with ASD and ID, and whether behavioural indicators will make it more easily identified. This population has been under-represented in research, and without cohort-appropriate anxiety measurement tools available, the true rate of anxiety comorbidity is unknown. As a result, individuals with ASD and co-occurring ID are at even greater risk of not receiving adequate services and supports and of their anxiety going untreated.

Anxiety comorbidity rates with ASD are consistently high, with research estimating an average rate of 40-42% (Hollocks et al., 2019; Lai et al., 2019; Simonoff et al., 2008). Psychiatric comorbidity in ASD is associated with poorer outcomes for individuals (Gillberg et al., 2016) and a great burden of care for families and the broader community (Baxter et al., 2015). There is a paucity of research available into how anxiety is identified and affects those with ASD and ID specifically, though the particular difficulties with diagnosing this population would suggest that the true rate of anxiety comorbidity for these individuals is currently under-reported. As a result, there is also a lack of treatment options, leaving individuals with ASD and ID at risk of going untreated for a significant mental health concern.

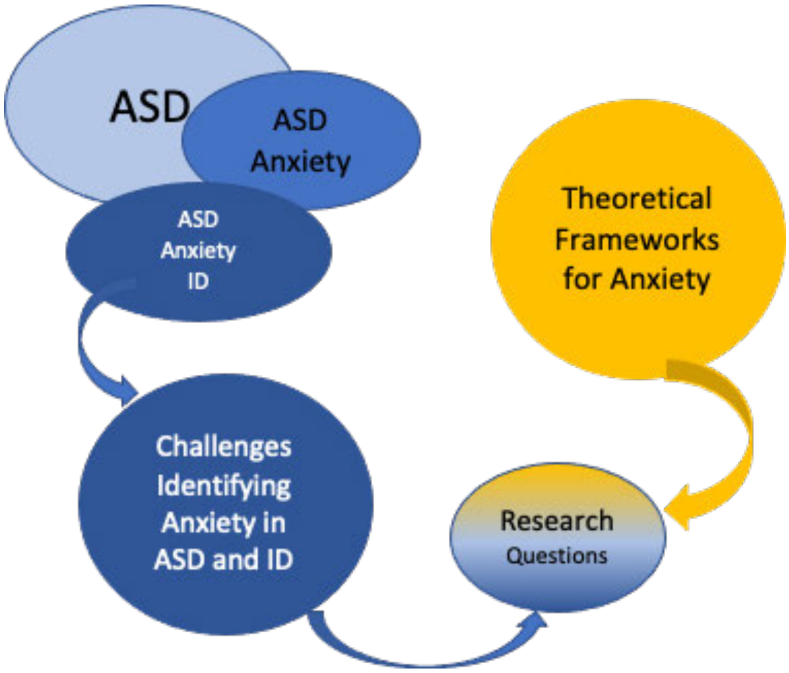
There is a dearth of specifically standardised anxiety measurement tools for use with individuals with ASD and ID. Without this key component for both research and clinical practice, it is unclear if anxiety is being consistently identified for this unique population. The use of behavioural markers as symptoms of anxiety offers an alternative pathway to identification and diagnosis. Problem or challenging behaviour is a possible indicator of elevated anxiety from existing research, with behavioural escalations being easily identified by carers of an individual with ASD and ID, and generally being attributed to their existing diagnoses. Challenging behaviours are a considerable issue for those with ASD and ID, with 82% reported to have at least one challenging behaviour; 96% more than one (Murphy et al., 2009). Podlesnik and Kelley (2017) report that challenging behaviours are persistent and often relapse or reinstate despite intensive behavioural intervention. By treating the underlying cause of problem behaviour (i.e. treatment of anxiety), the potential result is more significant behaviour change, which has to date been elusive. This in turn could result in individuals who are more available for learning and engagement, and require less intensive supports as a result of decreasing their anxiety and therefore their challenging behaviour. This would also reduce the burden and cost of care and increase quality of life.

Overview of the Thesis

The literature review, presented visually in Figure 1, draws together existing research in the areas of ASD, anxiety, and ID, with particular focus on the way these three conditions intersect in some individuals (Chapter 2). The role of ASD characteristics in this interaction is specifically considered. The challenges in identifying anxiety in those with ASD and ID are outlined in Chapter 3, including diagnostic categorisations and practices. This is addressed in a published scoping review of the existing research addressing this topic. Theoretical frameworks for anxiety development and particular vulnerabilities of those with ASD are then considered, with theories of anxiety discussed based on both neurotypical and ASD populations. These theoretical frameworks are presented in Chapter 4.

Figure 1

Literature Review Outline



The research questions are then presented, considering both the difficulties of anxiety diagnosis and theoretical framework for those with co-occurring ASD and ID (Figure 1). Two studies were completed as part of this research to answer these questions, with the results presented in Chapters 5 and 6. A general discussion considering the findings from both studies, the clinical implications, future directions, and conclusion are presented in Chapter 7.

Chapter 2: Autism, Anxiety, and Intellectual Disability

In this chapter, ASD and its characteristics will be reviewed. The nature of ASD characteristics and how they intersect with anxiety will be considered, with existing research presented. Comorbidity between ASD and ID with particular focus on research into anxiety within this population will then be outlined. As part of this thesis a scoping review of current research was published and is included in the next chapter.

Autism Spectrum Disorder

ASD is a neurodevelopmental disorder, characterised primarily by differences in social communication and RRBs. ASD is evident from early childhood, persisting across an individual's lifespan. These two clusters of core characteristics are both required for a diagnosis of ASD to be made; however, individuals can be variably affected from a mild to a severe degree for each cluster.

Core Characteristics of ASD

Social communication in ASD, as defined by the DSM-5 (APA, 2013), includes deficits in social-emotional reciprocity such as reduced interest in sharing emotions, interests, and experiences, failure to engage in back-and-forth conversation, and lack of initiation of or odd approach to social interactions. Deficits are also present in nonverbal communicative behaviours, indicated by a lack of ability, or limited ability, to coordinate nonverbal behaviours to communicate or enhance a message, such as using gestures and eye contact, with or without language. Difficulties are also noted in the development and maintenance of relationships, evident in difficulties in making and maintaining friendships or sharing imaginative play and in a lack of flexibility in altering behaviour to fit different situations.

RRBs are a core feature of ASD and can be a major barrier to engagement and social interactions (Leekam et al., 2011). Restricted behaviours and interests manifest in the inflexibility and perseveration of ideas and activities, intense focus, and desire for sameness in the environment. Repetition is seen in rhythmic motor movements and repetitive routines, language, and rituals (Leekam et al., 2011). Adults with ASD tend to show less repetitive behaviour than children with ASD, although this reduction over time is less pronounced for individuals with ASD and ID (Esbensen et al., 2009). The impact of RRBs can be considerable, as they can interfere with all aspects of development and functioning in children, including the development of social communication skills (Richler et al., 2010). Cuccaro et al. (2003) found that RRBs could be classified in one of two categories: *sensory motor* and *insistence on sameness*. Sensory motor includes behaviours such as repetitive use of objects, unusual sensory interest, complex mannerisms, and unusual preoccupations. Insistence on sameness describes difficulty with changes in routine, resistance to trivial changes, unusual response to sensory stimuli, compulsions/rituals, and sound sensitivity.

Information Processing in ASD

In addition to the core characteristics of autism, associated difficulties with information processing are common: theory of mind issues, weak central coherence, and executive dysfunction. Executive dysfunction in individuals with ASD leads to impairments in problem solving, strategic planning, and organisation, resulting in cognitive rigidity (Rajendran & Mitchell, 2007). That is, individuals with ASD can perseverate on details in a way that impedes solving or understanding the larger issue, leading to difficulties coping with change or solving problems as they arise. This can also be considered as cognitive inflexibility; individuals with ASD can find it challenging to switch between concepts or behaviours (Jiujias et al., 2017).

Central coherence refers to an individual's ability to see the "big picture" and therefore to understand how day-to-day actions play a part in a larger gestalt (Frith & Happé, 1994). Individuals with ASD tend to focus on details and fail to see the larger picture. Weak central coherence may also result in difficulties with generalising learning or skill acquisition across environments, as learning is context bound (de Marchena et al, 2015). This can make it difficult to predict future events and to extrapolate from previous experiences.

Theory of mind is part of typical development, whereby children learn that those around them are separate entities with their own thoughts, intentions, and emotions. Individuals with ASD are likely to struggle to attribute mental states to themselves and to others around them, making it difficult to infer what someone else believes and therefore predict how they will act (Baron-Cohen et al., 1985). This difficulty in predicting the actions and intents of others impacts social interactions and, it is also believed, an individual's ability to engage in imaginative play, as both require the ability to engage in second-order representation (Leslie, 1987).

Changes in the Diagnostic Process

The new conceptualisation of ASD in the DSM-5 aimed to unify the three previous diagnoses made under DSM-IV (APA, 1994) and DSM-IV-TR (APA, 2000); namely, autistic disorder, Asperger's syndrome, and pervasive developmental delay not otherwise specified (PDD-NOS). Therefore, based on when research was published, the terminology and diagnostic categories used to refer to autism may vary.

The DSM-IV diagnostic criteria for autistic disorder, published in 1994, described autism as a triad: communication/language delay before the age of 3, social reciprocity difficulties, and the presence of restricted and repetitive behaviours and interests. In the DSM-5 this has been reformulated, with social interactions and communication being combined to form a single cluster of social communication characteristics. The second cluster of characteristics, RRBs, was expanded to include sensory hypo- and hyper-sensitivities, sensory motor behaviours, insistence on sameness, and highly fixated interests. Each of these characteristics is then rated using descriptive severity levels of 1–3, with 3 indicating the highest level of support needed.

Another addition to the DSM-5 (APA, 2013) ASD diagnostic criteria is the inclusion of further specifiers. Comorbid diagnoses can now be included, in contrast to the DSM-IV-TR (APA, 2000), which explicitly excluded many co-occurring conditions such as attention deficit and hyperactivity disorder (ADHD); that is, one individual could not be diagnosed with both ADHD and autism. Specifiers include co-occurrence of ID, language impairment, associated medical conditions, and associated disorders. In this way, individuals with ASD can now have a comorbid diagnosis of generalised anxiety and/or ADHD, which were mutually exclusive conditions under DSM-IV.

Prevalence of ASD

The prevalence of diagnosed ASD has been steadily increasing over the last 20 years, doubling from an average occurrence in 2000 of 50/10,000 to an international average of 100/10,000 in 2022 (Zeidan et al., 2022). Zeidan et al. (2022) reported that incidence rates varied considerably internationally. For example, in America, the Centre for Disease Control and Prevention reported an average incidence of 185/10,000 in 8-year-olds (Baio et al., 2018); Y. Kim et al. (2011) reported an incidence of 264/10,000 in South Korea; a recent survey of children in Australia has reported an occurrence of 440/10,000 (May et al., 2020).

This steady increase can be attributed to several causes, including increased accuracy in screening for ASD (Smiley et al., 2018) and changing data collection methods and diagnostic standards (Fombonne, 2018). Research conducted into ASD diagnosis and prevalence prior to the DSM-IV publication considered only the more severe manifestations of ASD, such as autistic disorder (APA, 1994), without considering the broader autism spectrum encapsulated in the DSM-5 (APA, 2013). This, along with more complex data collection methods in epidemiological studies that capture information across a wider range of symptoms and characteristics, has had an impact on reported ASD rates (Fombonne, 2018). Differential diagnosis and accurate identification of ASD when it appears as a complex array of behaviours and symptoms has also had an impact on diagnostic rates. For example, a non-verbal individual with ASD, ID, and/or complicated mental health concerns was less likely to be diagnosed with autism because of the complex pattern of characteristics (Mohapatra et al., 2017). This increase in broader understanding of ASD has resulted in improved recognition, diagnostic process, and screening, now possible from 18 months on standardised testing (Myers et al., 2019).

ASD and Comorbidity: Anxiety

Individuals with ASD often experience and present with behavioural and emotional challenges in addition to the core features of their primary diagnosis. These behaviours can be challenging in nature and can have a significant impact on the development and learning of the individual with ASD, as well as increasing the burden of care on their family and support network (White et al., 2009). Examples of challenging behaviours include self-injurious behaviour (SIB), aggression, stereotypy, and other destructive behaviours (McTiernan et al., 2011). The frequency and intensity of challenging behaviours are positively correlated with ASD symptom severity (Matson et al., 2008) and level of ID (McTiernan et al., 2011).

Research into children with autistic disorder and PDD-NOS and using the DSM-III-R (APA, 1987) anxiety criteria found that, overall, 84% of the children met criteria for a clinically significant anxiety disorder (Muris et al., 1998). The most common anxiety disorders were simple phobias, though the researchers noted that several may be present, followed by separation anxiety and then avoidant disorder.

Complex behaviours and the lifelong nature of ASD are thought to contribute to a higher risk of developing a comorbid psychiatric disorder within this population. Leyfer et al. (2006) found that in their sample of children with ASD, 72% met criteria for a comorbid psychiatric diagnosis using DSM-IV (APA, 1994); of those, 50% met criteria for two or more psychiatric disorders. The most common of these were specific phobia (44%), obsessive compulsive disorder (OCD; 37%), and ADHD (30%).

Similar results were reported by Simonoff et al. in 2008; 70.8% of their participants met criteria for a diagnosis secondary to ASD, with 41% of those meeting criteria for two or more disorders (again using DSM-IV criteria). In this study the most common comorbid disorders were social anxiety disorder (29.2%), ADHD (28.1%), and oppositional defiant disorder (28.1%). More recently, in a meta-analysis, Lai et al. (2019) found that those with ASD were significantly more likely to meet criteria for co-occurring psychopathology with ADHD (33%), anxiety disorders (23%), and depressive disorders (12%) being the most common.

Consistently, research is finding that a diagnosis of ASD increases an individual's risk of a comorbid psychiatric condition significantly. It is also clear that this co-occurring psychiatric condition is highly likely to be an anxiety disorder or phobia, which is classified as an anxiety disorder in both DSM-IV and DSM-5 (APA, 1994 and APA, 2013).

Anxiety has long been connected with ASD. Kanner stated in 1943 that one of the discriminating criteria for autism was the presence of anxiously obsessive behaviour patterns that ensured sameness and predictability for an individual, with rage or panic being the result when these strategies failed. Groden et al. (2006) likewise noted that individuals with ASD are likely to use behavioural means to maintain their emotional stability and internal homeostasis, particularly when experiencing stressors such as unpredictable change. Examples of such behavioural mechanisms are the creation of routines and use of repetitive behaviours in order to ensure consistency and predictability.

Defining anxiety is complex, with DSM-5 providing diagnostic criteria for 10 distinguishable anxiety-based disorders (APA, 2013). Research has focused on both individual anxiety disorders and all anxiety disorders as a combined group when investigating and reporting on anxiety rates and diagnostic processes. Here, anxiety will be considered in general terms which align with the definition of generalised anxiety disorder (GAD). As such, anxiety can be defined as an internal and chronic state resulting from persistent stress exhibited by excessive anxiety or worry about a number of events or activities, which is not easily controlled and is evident through an array of physiological changes (DSM-5; APA, 2013). Physiological or physical experiences associated with anxiety can include muscle tension, restlessness, being easily fatigued, sleep disturbance, accelerated heart rate, and dizziness. Both cognitive and physiological aspects need to be present for a diagnosis of anxiety to be made.

As mentioned previously, comorbid rates of ASD and an anxiety disorder average around 40-42% (Hollocks et al., 2019; Lai et al., 2019; Simonoff et al., 2008). These researchers have studied individuals with ASD who also meet full criteria for an anxiety disorder at the time of testing or in the previous 3 months. What is not captured is the presence of subclinical anxiety; this would represent subthreshold symptoms indicating increased susceptibility to developing an anxiety disorder, or the partial remittance of a previously clinical level disorder (Vasa et al., 2013). Research specifically into subclinical levels of anxiety in children with ASD has found results ranging from 20% (Niditch et al., 2012) to 26% (Vasa et al., 2013). This could then represent an additional proportion to the 40% of those meeting full criteria for clinical anxiety disorder. Thus, anxiety, either at a clinical or subclinical level, represents a significant risk and therefore a concern for those with ASD.

With anxiety representing a significant concern for those with ASD, it is unsurprising that research has started to focus on this relationship (Lai et al., 2019). In attempting to investigate the specific vulnerability that ASD presents in relation to anxiety, it is useful to consider how the core characteristics, information processing, and other associated correlates of ASD impact this relationship. As such, the next section will discuss the available research into ways individual characteristics of ASD have been found to be associated with anxiety.

Communication and Anxiety

Individuals with ASD and higher communication skills are more likely to experience anxiety than those with ASD and lower communication skills (Davis et al., 2012; Dickerson Mayes et al., 2011; Kerns & Kendall, 2013; Weisbrot et al., 2005; White et al., 2009). These findings suggest that individuals with more severe communication deficits actually experience less anxiety than those with ASD who have more verbal and communication abilities. It is possible that those with more severe communication deficits, which are associated with more cognitive impairments, may possess a decreased ability to generate and experience a complex emotion such as anxiety (Davis et al., 2012; Davis et al., 2011; Kerns & Kendall, 2013). Gobrial and Raghaven (2012) pointed out that a deficit in communication skills results in greater difficulties discussing and understanding fears, which can result in an overgeneralisation of fear to an environment or situation, and therefore anxiety. However, Tsai (1996) suggested that level of functioning affects the way anxiety is expressed, rather than what is experienced by the individual. Similarly, Davis et al. (2012) and Kerns and Kendall (2013) proposed that increased communication difficulties result in individuals with ASD being unable to report their experience of anxiety, rather than having decreased experiences of anxiety in general. Davis et al. (2011) also suggested that communication difficulties prevent children with ASD from expressing or showing symptoms of anxiety, as they are currently defined. They raise the possibility that some anxiety symptoms may be misinterpreted as communication deficits for individuals with ASD.

Social Functioning and Anxiety

Limited research has focused on the specific relationship between social abilities and anxieties within an ASD population. Chang et al. (2012) found that in children with ASD and a pre-existing anxiety disorder, increased severity of social anxiety was associated with a higher level of social functioning deficits. Bishop-Fitzpatrick et al. (2013) found, in adults with ASD and no co-occurring ID, that as carer reported stress increased, social functioning decreased. J. Green et al. (2000) studied the relationship between social functioning and psychiatric comorbidity in a group of individuals with Asperger's syndrome; individuals and their families reported severe social impairments and high levels of anxiety, although whether this can be extrapolated across the autism spectrum is unclear given the high cognitive abilities of the group. Mayes et al. (2011) mentioned that social avoidance for individuals with ASD may be motivated by social indifference or by a lack of skills, while social phobia results from social anxiety. It has also been noted that social skills training needs to be incorporated into cognitive behavioural therapy when treating anxiety for individuals with ASD (Sofronoff et al., 2005). However, this form of therapy is language intensive, and therefore inaccessible for many individuals with ASD, particularly those with ID.

Restricted and Repetitive Behaviours and Anxiety

The high comorbidity of ASD and anxiety could, at least in part, be explained by some similarities in diagnosis, such as the presence of RRBs. RRBs, as a core component of ASD, have been thought to be predominantly sensory or enjoyment based and research has suggested several motivations: pleasure (Dickerson Mayes et al., 2011), self-occupation (Leekam et al., 2011), sensory stimulation, and situation avoidance or attention seeking (Joosten et al., 2009; Militerni et al., 2002). However, it has also been suggested that RRBs have an anxiety management function for individuals with ASD as well (Cashin & Yorke, 2018; Joyce et al., 2017). Joosten et al.

(2009) found that in children with ASD and ID, anxiety was closely associated with and appeared to be a more likely underpinning for RRBs than sensory seeking. Davis et al. (2011) suggested that RRBs act as a soothing or coping strategy for children with ASD when they experience stress or anxiety. The behaviours may act as a distraction or for comfort during high-stress situations, particularly in the absence of other coping mechanisms such as avoidance or escape (Davis et al., 2010). Rodgers et al. (2012) suggested that both sensory motor and insistence on sameness (IS) behaviours may work as a short-term alleviation of stress by reducing the demands and restricting the environment, thereby giving the individual with autism control. This pattern may then become reinforced through positive experience and limit the development of other strategies to cope with stress and anxiety.

Gotham et al. (2013) noted that IS behaviours were remarkably close to anxiety behaviours and postulated that this explained the high comorbidity of ASD and anxiety. They proposed a model to test the theory that IS and anxiety were separate constructs, as opposed to anxiety being a component of ASD via IS. They concluded that IS and anxiety were relatively independent, and anxiety was therefore a separate construct to ASD.

Information Processing and Anxiety

No research could be found that specifically considered executive functioning or central coherence difficulties in relation to anxiety. A single study could be found that considered how theory of mind related to anxiety: Hunsche et al. (2022) found that children with ASD who were socially anxious were also likely to have impaired theory of mind and in turn to present with distinct social fears. The authors suggested that specific ASD cognitive features, such as theory of mind difficulties, could play a role in the distinct vulnerability of individuals with ASD in terms of developing anxiety. Lainhart (1999) mentioned that the nature of executive dysfunction could increase an individual's inability to cope with change, as well as causing a decrease in effective problem solving when faced with stressful situations, therefore increasing anxiety. Jiujias et al. (2017) pointed out that cognitive flexibility deficits have been found to correlate with higher levels of RRBs. Via this pathway, specific components to executive functioning may be associated with increased anxiety. Weak central coherence diminishes the individual's ability to predict the world and people around them, which could then result in severe panic, fear, and anxiety (Lainhart, 1999). Theory of mind difficulties can lead to difficulty in reading social intent and to breakdown in communication with others, which in turn results in stress and, longer term, in anxiety (Lainhart, 1999).

Emotional Regulation and Anxiety

Another factor that may influence an individual with ASD's vulnerability to anxiety is emotional dysregulation. This is the failure to modulate the intensity or duration of emotions or motivations to achieve goals or social adaptation (Jahromi et al., 2012). Emotional regulation is generally learnt in early childhood and enables children to adapt to and cope with negativity and frustrations by learning appropriate coping strategies. Dysregulation results in disruptive behaviours such as irritability, tantrums/meltdowns, self-injurious behaviour, aggression, poor anger control, and mood swings (Samson et al., 2014), and can moderate developmental trajectories and increase negative outcomes for children (Jahromi et al., 2012). Though not considered a core feature of ASD, there is growing research evidence that emotional dysregulation is a serious concern for individuals with ASD (Prizant et al., 2003).

Emotional dysregulation has been linked to executive functioning through internal deficits in self-monitoring (Jahromi et al., 2012), resulting in individuals with ASD being more vulnerable to regulation difficulties. Samson et al. (2014) reported that there is a significant relationship between emotional dysregulation and ASD symptomology, with RRB severity being the best predictor of emotional dysregulation. Samson et al. (2015) later found that individuals with ASD more frequently used maladaptive emotional regulation techniques when experiencing anxiety or anger than their non-ASD peers. They were more likely to utilise RRBs as a coping mechanism, rather than strategies that required higher executive functioning such as problem solving or cognitive reappraisal. Gotham et al. (2015) also found that in individuals with ASD and non-spectrum developmental delay, with mixed cognitive abilities, higher levels of anxiety symptoms were associated with more impaired emotional regulation.

Sensory Sensitivities and Anxiety

The presence of hyper- or hypo-reactivity to sensory input is listed as a diagnostic feature for ASD in the DSM-5. This refers to limited or no response to sensory input (under-responsivity/hyporeactivity) or exaggerated, rapid onset and/or prolonged response to sensations (over-responsivity/hyperactivity; Ben-Sasson et al., 2008). Sensory over-responsivity (SOR) is associated with negative emotionality, anxiety, withdrawal, and separation distress, and leads to further functional and social impairments (Ben-Sasson et al., 2008). SOR has been found to often co-occur with elevated anxiety symptoms in individuals with ASD (Ben-Sasson et al., 2009; Black et al., 2017; Liss et al., 2008). S. Green et al. (2012) found that SOR was evident before anxiety, and therefore postulated that SOR may exacerbate a predisposition to anxiety, or that SOR may increase the risk of developing anxiety. The experience of SOR may become generalised to an environment or situation, particularly when a lack of other coping strategies exists, resulting in hypervigilance and anxiety (S. Green & Ben-Sasson, 2010).

Researchers have found variable results between the characteristics of ASD and presence of anxiety, with no clear and consistent relationship being found between a single characteristic and anxiety to fully explain the increased vulnerability. What is clear from the above research is the lack of clarity about the relationship between characteristics of ASD and symptoms of anxiety; as a result, it is not possible to definitively assign anxiety symptoms to a single characteristic. The very nature of ASD and its associated communication and information processing difficulties also hamper our ability to gather meaningful data, draw conclusions, or generalise findings across the autism spectrum.

ASD and Comorbidity: Intellectual Disability

Like ASD, the diagnosis for ID has been redefined with the publication of the DSM-5 in 2013. Previously, ID (labelled mental retardation) was considered in terms of intellectual functioning alone and traditionally was diagnosed when an individual's IQ was less than two standard deviations below the norm—that is, typically less than 70. Intellectual functioning includes abilities such as reasoning, problem solving, abstract thinking, and academic and experiential learning (APA, 2013). However, in the DSM-5 a diagnosis of ID requires the presence of deficits to both intellectual functioning and adaptive behaviours. Adaptive behaviours enable people to independently access and participate in the world around them. Deficits in adaptive abilities can have a significantly negative impact an individual's social participation, communication, personal independence, and community access. In terms of diagnosis, it is the level of adaptive ability rather than the IQ level that defines the level of ID diagnosed, as adaptive ability determines the level of support the individual will require in day-to-day life. This is particularly true at the lower end of the IQ range, as standardised IQ tests often have a basal score of 40, so differentiation between severe and profound levels of functioning can be difficult (Rivard et al., 2015).

The change in diagnostic process in the DSM-5 has a considerable impact on prevalence rates between ASD and ID. Under DSM-IV criteria, the comorbidity of autistic disorder and ID was close to 70% (Chakrabarti & Fombonne, 2005; Yeargin-

Allsopp et al., 2003). However, it was also noted that in considering the broader concept of pervasive developmental delay in the DSM-IV, those without an ID (Asperger's syndrome and PDD-NOS) made up the majority of diagnoses (70%; Chakrabarti & Fombonne, 2005). Zeidan et al. (2022), in a meta-analysis of research, found the average co-occurrence of 33% for ASD and ID across studies.

The interaction of ASD and ID can result in different behavioural profiles across levels of ID. Brereton et al. (2006) reported that children with ASD and a moderate-severe ID present with more self-absorbed and social relating problems when compared to those with ASD and a borderline or normal IQ. Militerni et al. (2002) reported that children with ASD and a lower IQ were more likely to engage in self-stimulatory activities than those with ASD and a higher IQ. Thus, the co-occurrence of ASD and ID seems to have an amplifying effect for ASD symptom severity, and as such presents differently from ASD or ID alone (Deb & Prasad, 1994). It also appears that the level of ID has a differential impact, meaning that individuals with ID do not present as a homogenous population.

The co-occurrence of ASD and ID also seems to increase an individual's vulnerability to developing behaviours that challenge, and perhaps to more complex psychopathology. For example, Murphy et al. (2009) found that as ASD symptom severity and level of ID increase, the likelihood of an individual engaging in challenging behaviours, particularly self-injury, also increases. Hill and Furniss (2006) reported that those with severe ID and ASD showed higher levels of anxiety, stereotypy, mania, and mood disturbance than those with severe ID alone. Bradley et al. (2004) found that individuals with ASD and ID were more likely to have a comorbid psychopathology than persons with ID alone.

Despite these findings, little research has focused on those with comorbid ASD and ID, particularly in terms of their mental health. This can partly be explained by difficulties in diagnosis. The inherent language and communication difficulties associated with ASD can be further exacerbated by the presence of an ID, limiting an individual's ability to understand and express themselves. Given that the presence of an ID seems to amplify ASD symptomology, it would be possible for severe ASD to overshadow the existence of an ID (Hill & Furniss, 2006). As ASD and ID together seem to present differently than either disability alone, it seems clear that specific diagnostic scales are necessary when considering the possibility of comorbid psychopathology (Matson & Shoemaker, 2009).

ASD, ID, and Anxiety

Kerns and Kendall (2013) discussed the relationship between intellectual functioning and anxiety within an ASD population, citing an inconsistent array of results from research. Lecavalier (2006) found that higher cognitive abilities were associated with higher anxiety symptoms, as did Muris et al. (1998), Mayes et al. (2011), Sukhodolsky et al. (2008), Weisbrot et al. (2005), and Gadow et al. (2005). Muris et al. looked at children diagnosed with autistic disorder and PDD-NOS, noting those with autistic disorder having a significantly lower IQ, with a mean of 70. They found children with autistic disorder had lower levels of clinical anxiety disorders than their PDD-NOS peers, though they did show high levels of simple phobia and some separation anxiety, but not the avoidant or overanxious disorders of the children with PDD-NOS. These authors suggested that age and intellectual ability may play a role in some types of anxiety phenomena, making worry and fearful anticipation unavailable to some individuals. Sudhodolsky et al. specifically mentioned that while the occurrence of some anxiety disorders was affected by intellectual ability, such as generalised, separation, and panic related/worry, others (specific and social phobia) were consistently prevalent in children with ASD regardless of their intellectual ability. Likewise, Mayes et al. noted that children with ASD and higher IQs reported higher scores on anxiety symptoms such as worry, self-consciousness, and being sick with worry, but showed no difference from those with lower IQs for more behavioural symptoms such as oppositional behaviour and avoidance. These findings would suggest that intellectual ability may play a role in the manifestation of anxiety rather than its existence, while also highlighting the difficulties in operationalising anxiety for research for those without the language skills to describe their experience.

Scant research exists into the co-occurrence of ASD, ID, and anxiety. The co-occurrence of ID with ASD is an added complication that makes identifying anxiety increasingly difficult. The same issues exist for those with ASD and anxiety, in that communication limitations and lack of meaningful data collection hamper our ability to identify anxiety. The presence of ID with ASD only exacerbates these difficulties, making individuals in this population even harder to definitively identify and diagnose as experiencing anxiety. Considering these difficulties and how we can use other means to address these limitations will be considered in Chapter 3.

Chapter 3: Challenges in Identifying and Diagnosing Anxiety

In this chapter, research that directly addresses the way anxiety is identified and diagnosed in those with ASD and ID will be considered. A myriad of issues exist, including symptom amplification, diagnostic overshadowing, and the lack of measurement tools with appropriate normative data for this population. In a published scoping review completed as part of this thesis, only nine studies published between 1994 and 2019 were found that considered how anxiety can be identified in those with ASD and ID. The published paper is included here, having first been published in its entirety in the *Journal of Mental Health Research in Intellectual Disability*. The paper has been reformatted to allow for continuity here:

Appleton, H., Roberts, J., & Simpson, K. (2019). How is anxiety identified and diagnosed in individuals with autism spectrum disorder and intellectual disability? A scoping review. *Journal of Mental Health Research in Intellectual Disability.* https://doi.org/10.1080/19315864.2019.1679299

Abstract

Introduction: Individuals with Autism Spectrum Disorder display a pattern of social communication deficits and restricted and repetitive behaviours that leave them particularly vulnerable to developing anxiety. The presence of a co-occurring Intellectual Disability further complicates the situation, compromising traditional diagnostic techniques and processes. The dual diagnosis of ASD and ID appears to result in specific behavioural patterns that affect the way anxiety is identified in this population.

Method: A scoping review was undertaken to explore what is currently known about the way anxiety is identified and diagnosed in individuals with ASD and ID.

Results: In the limited research available consistent themes of difficulties with the diagnostic process, inconsistencies among measurement tools and the need to consider behavioural symptomology were found.

Conclusion: Further research needs to be conducted to enhance our understanding of how anxiety is identified in those with ASD and ID. This research could more accurately inform reliable diagnostic processes and lead to better treatment and outcomes for this population.

Keywords: autism spectrum disorder; intellectual disability; anxiety.

How is Anxiety Identified and Diagnosed in Individuals with Autism Spectrum Disorder and Intellectual Disability: A Scoping Review

Children and young adults with Autism Spectrum Disorder (ASD) are well documented to experience high rates of co-occurring anxiety disorders, with prevalence rates varying between 42% (Bradley et al., 2004) and 80% (Simonoff et al., 2008). The presence of a co-occurring Intellectual Disability (ID) is also relatively common in ASD, with rates reported to be 37% (Rivard et al., 2015). While individuals with ASD and ID make up a significant proportion of the ASD population, research into the way anxiety affects these individuals is scant and divergent. For example, Lecavalier (2006) suggests this group experiences significantly less anxiety than individuals with ASD and no ID, while Hill & Furniss, (2006) suggest that individuals with ASD and ID are more at risk of developing anxiety than those with ASD alone.

ASD and Anxiety

ASD is a neurodevelopmental disorder, characterised by differences in social communication and restricted and repetitive behaviours. It is evident from early childhood and persists across an individual's lifespan (Diagnostic and Statistical Manual

5th Edition (DSM-5): American Psychiatric Association (APA), 2013). These two clusters of core characteristics are both required for a diagnosis of ASD. An individual's functioning can be variably affected from a mild to a severe degree for each cluster.

Defining anxiety is a more complex process, with DSM-5 providing diagnostic criteria for ten distinguishable anxiety-based disorders (APA, 2013). Research has focused on both individual anxiety disorders and all anxiety disorders as a combined group when investigating and reporting on anxiety rates and diagnostic processes. For the purposes of this paper, anxiety will be considered in its most general terms to allow for the broadest application, which most strongly aligns with the definition of Generalised Anxiety Disorder (GAD). As such, anxiety can be defined as an internal and chronic state resulting from persistent stress exhibited by excessive anxiety or worry about a number of events or activities, which is not easily controlled and is evident through an array of physiological changes (DSM-5: APA, 2013). Physiological or physical experiences associated with anxiety can include muscle tension, restlessness, being easily fatigued, sleep disturbance, accelerated heart rate and dizziness. Both cognitive and physiological aspects need to be present for a diagnosis of anxiety to be made.

The connection between ASD and anxiety is long standing; one of Kanner's discriminating criteria for autism in 1943 was the presence of anxiously obsessive behaviour patterns that ensured sameness and predictability for an individual, with rage or panic being the result when these strategies failed. Groden et al. (2006) likewise note that individuals with ASD are likely to use behavioural means to maintain their emotional stability and internal homeostasis, particularly when experiencing stressors such as unpredictable change. Examples of such behavioural mechanisms are the creation of routines and use of repetitive behaviours in order to ensure consistency and predictability. Diagnosis of anxiety in individuals with ASD has typically followed the same process as for their neurotypical peers. That is, through clinical interviews with medical or psychiatric specialists, or, particularly for research purposes, using self or carer completed psychopathology or anxiety measurement tools (Grondhuis & Aman, 2012). The focus of these tools, and the diagnostic criteria used to identify anxiety, has been the identification of a unique symptom subset that can be used to differentiate anxiety from ASD (Helverschou et al., 2008). However, there are striking similarities between the behaviours exhibited by those with ASD and those with a diagnosed anxiety disorder; for example, behaviours such as: social withdrawal, repetitive and restricted behaviours, ritualistic or compulsive behaviours and atypical attention (Morgan, 2006). This difficulty with delineation has led some researchers to suggest that rather than a separate set of behaviours, comorbid anxiety could be identified through ASD symptom amplification (Kerns et al., 2014), such as increased repetitive behaviours (Rodgers et al., 2012; Spiker et al., 2011).

Kerns et al. (2017) particularly note not only the diagnostic overlaps in symptomology between ASD and anxiety, but also the ambiguous anxiety-like symptoms that are specific to those with ASD. They particularly note specific sources of anxiety for individuals with ASD such as changes in routine, anticipation of change, anxiety around a lack of clear social rules and expectations and intense fears of odd items/experiences or specific phobias. There is a lack of clarity around where these ASD specific experiences belong diagnostically, as they describe both ASD and anxiety experiences. Kerns et al. have tried to encapsulate this difficulty with adaptions to the Anxiety Disorders Interview Schedule-Child/Parent: Parent version (Silverman & Albano, 1996), namely the Autism Spectrum Addendum (ADIS/ASA) (Kerns et al., 2014). Research into the use of this measure has found it useful to consider both the traditional anxiety diagnostic categories and the ASD specific ambiguous anxiety symptoms when considering the comorbidity of ASD and anxiety (Kerns et al., 2017).

ID and ASD

Intellectual Disability (ID) is currently defined in DSM-5 (APA, 2013) as a significant deficit in both cognitive functioning and adaptive behaviours, with the degree of disability based more on adaptive behaviour assessment and the assessed need for supports in day-to-day life rather than IQ. Individuals are diagnosed as having a mild, moderate, severe or profound intellectual disability.

Individuals with ID have been found to be more susceptible to developing a wide array of psychopathology, anxiety included, than their non-ID peers (Pyles et al., 1997). Researchers have reported that severity of ID appears to affect this prevalence rate, with several studies finding that individuals with a severe ID experienced lower rates of anxiety than those with a mild or moderate ID (Einfeld & Tonge, 1996; Holden & Gitlesen, 2004). However, diagnosing anxiety in individuals with ID faces similar difficulties to those for individuals with ASD, particularly as the severity of ID increases and self-report becomes less accessible (Bailey & Andrews, 2003). Several ID specific anxiety measures have been developed in an attempt to address these diagnostic difficulties, however in a review of 14 such measures, Hermans et al. (2011) found that few of the developed measures had good reliability or consistency.

The interaction of ASD and ID can result in different behavioural profiles related to the severity of ID. Brereton et al. (2006) report that children with ASD and a moderate-severe ID present with more self-absorbed and social relating deficits when compared to those with ASD and a borderline or typical range IQ. Militerni et al. (2002) report that children with ASD and a lower IQ were more likely to engage in self-stimulatory activities than those with ASD and a higher IQ. Therefore the co-occurrence of ASD and ID could have an amplifying effect for ASD symptom severity, and as such presents differently from ASD or ID alone (Deb & Prasad, 1994). It also appears that the level of

ID has a differential impact, meaning that individuals with ID do not present as a homogenous population.

The co-occurrence of ASD and ID appears to create a specific vulnerability to developing difficult behaviours and psychopathology. For example, Murphy et al. (2009) found that as ASD symptom severity and level of ID increases, the likelihood of an individual engaging in challenging behaviours also increases, particularly self-injury. While Hill and Furniss (2006) report that individuals with severe ASD and ID showed higher levels of anxiety, mania and mood disturbance compared to those with severe ID alone. Despite these findings, little research has focused on this group and their mental health.

ASD, ID and Anxiety. Definitions of anxiety include a combination of physiological and cognitive aspects, highlighting the physical changes someone with anxiety will experience because of excessive worry about possible future events (APA, 2013). Communication difficulties are associated with both ASD and significant levels of ID and potentially have a considerable impact on the diagnostic process. Clinical interviews are language based by definition and optimal diagnostic and treatment outcomes require the individual to be able to express their internal state accurately. Lainhart and Folstein (1994) point out that individuals with ASD can find abstract thinking difficult, which in turn affects their understanding of internal concepts and the impact of their mood and emotions. The traditional interview process and completion of self-report questionnaires is vulnerable to difficulties in both receptive and expressive communication, resulting in only limited information for professional decision-making (Grondhuis & Aman, 2012). Diagnosis is therefore reliant on caregiver feedback or completion of questionnaires. This information gathering relies on caregivers being able to accurately interpret the behaviour of their child or client with ASD and ID, and accurately report this back. However, Helverschou and Martinsen (2011), found that caregivers often failed to identify physiological markers in those they cared for, making reporting anxiety related behaviours and therefore diagnosis, difficult.

MacNeil et al. (2009) completed a literature review into the diagnosis of anxiety in children and adolescents with ASD. In this review they found that the most reliable diagnostic process was multimodal; that is, multiple informants were used, multiple assessment techniques (clinical interview, observation, physiological assessment), in addition to ASD appropriate standardised measurement tools. This review did not specifically look at individuals with ASD and ID, but offers direction for further research, as it addresses many of the concerns raised about current diagnostic processes.

The limitations of the diagnostic process for this population, may therefore result in an underestimation of the comorbidity of ASD, ID and anxiety. In considering a more effective diagnostic protocol, Tantam (2000) suggests that observable behavioural patterns and the presence of atypical behaviours for the individual may represent a more reliable means of diagnosing anxiety in individuals with ASD and ID. It is possible that a behavioural pattern exists that better identifies anxiety in this population, leading to more reliable diagnosis and therefore development of more effective treatment and improved outcomes.

Research Question: What is known about how anxiety is identified and diagnosed in individuals with ASD and ID?

Method

A scoping review was undertaken to investigate the current literature addressing the way anxiety is identified and/or diagnosed in individuals with ASD and ID. The aim of scoping reviews is the synthesis of information about a topic to allow for key concepts and types of evidence to be mapped, to identify gaps in our knowledge and set the direction for future research, intervention development and increased community understanding (Colquhoun et al., 2014). Given the emerging nature of the research into ASD, ID and anxiety, a review of the current literature across a range of disciplines and methodologies was undertaken to explore the relevant research and to identify existing gaps.

Search Procedure

A search of the available literature was conducted through PsychInfo, Proquest and Medline, published between 1994 to present day. This start date was used as it was when DSM-IV was published and the diagnostic criteria for autism/autistic disorder substantially developed. Table 1 details the search terms used in order to garner the largest field possible and to capture the intersection of the three elements of interest: ASD, ID and anxiety. As a result, "autis*" was used to cover a wide variety of labels for ASD. Asperger's Syndrome was not included in this search given that this group, by definition, does not have a co-occurring ID. Internationally different terms are used in reference to ID and this terminology has changed with time, therefore a range of terms were used to identify relevant papers. Individual specific anxiety disorders were included as well as the more general term "anxiety", to maximise the research papers captured by the search.

Table 1

Search terms used for scoping review of ASD, ID and anxiety

ASD	ID	Anxiety
Autis*	Intellectual Disability	Anxiety
	Intellectual Delay	Generalised Anxiety Disorder
	Developmental Disability	Social Phobia
	Developmental Delay	Social Anxiety Disorder
	Learning Disability	Panic Disorder

Mental Retardation	Obsessive Compulsive Disorder
	Posttraumatic Stress Disorder
	Agoraphobia
	Specific Phobia
	Separation Anxiety

The search included Title, Abstract or Keyword search fields. All searches included an identifier of ASD and at least one for ID and for anxiety. The Refinements added to the search were for articles that had Full Text available, were Human in nature and were available in English. During the Proquest search, the article search was also refined to include Scholarly Journals, Dissertations & Theses, while newspapers, trade journals, wire feeds and magazines were excluded. For thoroughness, the reference list for each of the identified papers was also reviewed. The same process was followed as above for identifying papers of interest; titles were searched for an identifier of ASD, at least one for ID and anxiety.

Further exclusions were then applied after reading the abstracts. The following were removed; literature review papers, family-based studies (such as those focusing on the mental health of parents, siblings or carers), mice gene studies, journal information (such as reviews, opinions, brief overviews and errata notices), other specific disabilities as the main interest (such as Fragile X and Retts) and papers that did not include participants with combined ASD, ID and anxiety symptomology. The selected papers were read in full and included in the final review if they addressed the way anxiety was identified and diagnosed in individuals with ASD and ID.

Two reviewers assessed the papers independently to ensure reliability. For this process, 25% of the titles and abstracts of these papers were read by a second reviewer and following the same exclusion criteria, an agreement of 87% was reached. A third reviewer then read the selected papers in full to ensure final inclusion/exclusion criteria were agreed upon, resulting in an initial agreement of 90.6%. After discussion between the reviewers of the inclusion and exclusion criteria, an agreement of 100% was reached for the selected papers.

Analysis

The selected studies were evaluated using the same two-step process as MacNeil et al. (2009) used in their review of anxiety in children and adolescents with ASD. The first step was to consider the recommendations from research about how anxiety should be assessed and diagnosed for this population, isolating six key components for accurate assessment. Each paper was rated as “yes=1” or “no=0”, with a total possible score of 6.

(1) Use of clinical interview
(2) Use of anxiety rating scales
(3) Use of direct observation
(4) Use of physiological measures
(5) Use of multiple informants
(6) Use of assessment methods that have comparison data available for ASD samples.

A second evaluation was undertaken to focus on the methodological quality of the papers under review. MacNeil et al. (2009) modified Boyle’s 1998 checklist for prevalence studies and included the following components. Again, each paper was rated as “yes=1” or “no=0”, with a total possible score of 8.

(1) Description of sample
(2) Confirmation of ASD diagnosis
(3) Inclusion of comparison group
(4) Matching of comparison groups

(5) Standardising the method of data collection

(6) Reliability of anxiety instruments

(7) Validity of anxiety instruments

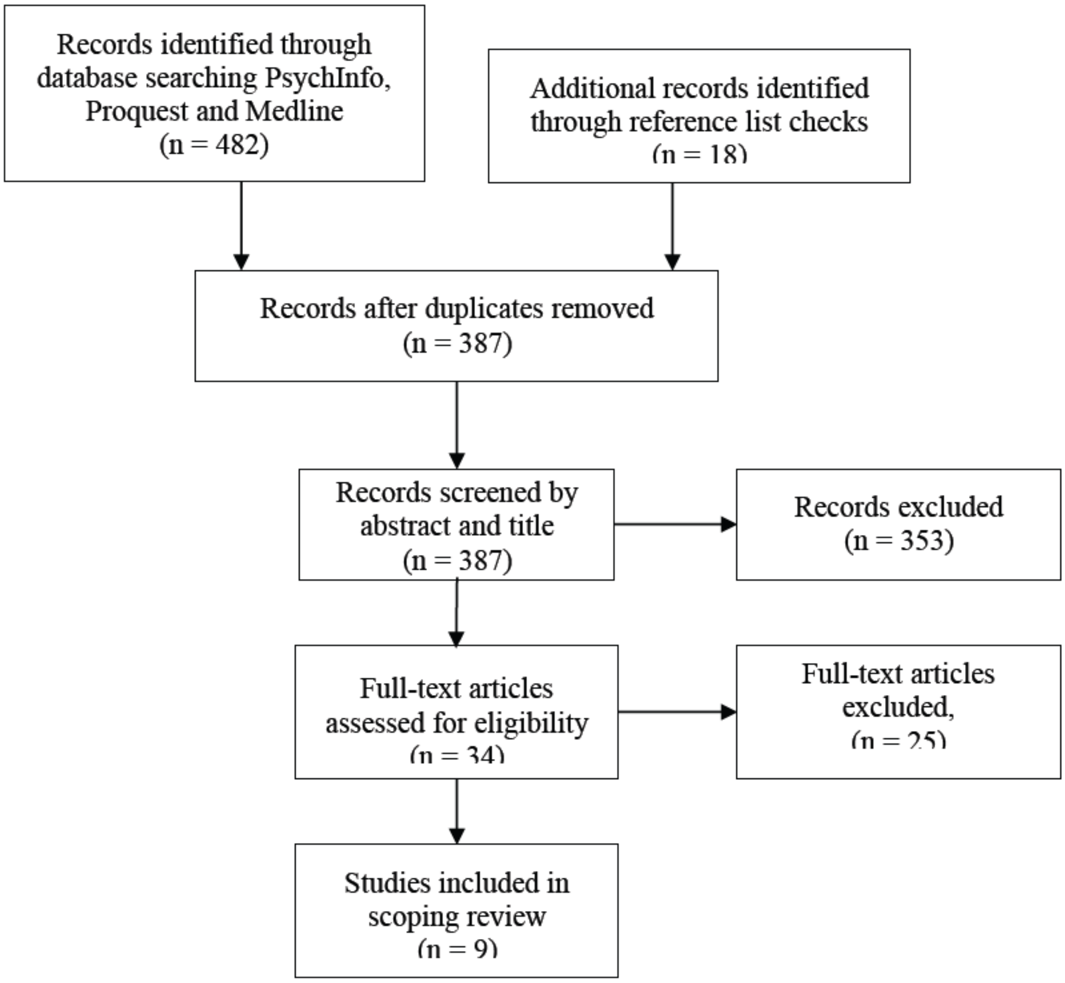
(8) Appropriateness of interpretations

Results

The initial search yielded 482 potentially relevant citations. After duplications were removed, 387 citations were examined to determine if they met the eligibility criteria based on title, abstract and key word search, resulting in 34 papers being identified. These papers were read in full, with those not addressing how anxiety was measured or identified in individuals with both ASD and ID being excluded, resulting in 9 papers for final inclusion. The selection process is charted in Figure 2.

Figure 2

Flow chat of article selection process



The general characteristics of the nine studies meeting the eligibility criteria are summarised in Table 2. Across the nine papers, there were 764 participants, aged between 3 and 57 years old. There was variability in the number of participants in the studies, ranging from three to 313. Participants with an ID all had an IQ < 70, and all levels of ID were grouped together in the reporting of results. In some of the studies a comparison group of participants with ASD and no ID was included; for these participants their IQ was reported as >70. Participants were drawn from disability specific services, including health services and special schools. Only two studies reported prevalence rates for anxiety co-occurring with ASD and ID: 37% by Helverschou and Martinsen (2011) and 43% by Sukhodolsky et al. (2008).

Table 1

Studies examining the identification of anxiety in individuals with ASD and ID

Researchers	Participants	Comparison group	Participants drawn from	Anxiety tool / Diagnosis method	How anxiety defined	Outcomes
Helverschou, S. B., Bakken, T. L., & Martinsen, H. (2008)	9 clinicians	n/a	2 specialist health services	Symptoms drawn from ICD-10 & DSM-IV; items for specific conditions gathered from a range of disability specific measures: PAS-ADD, PIMRA, DASH-II, Y-BOCS, GAF, PANSS, HAD, K-SADS, ABC, ADI-R	12 items covering *physiological arousal* and 4 for *avoidance*, 1 for *cognition*.	Anxiety disorders can be identified in individuals with ASD +ID with similar symptoms as typically developing peers More cognitive based criteria not clinically useful in diagnosis focus on physiological symptoms.
Helverschou, S. B., & Martinsen, H. (2011)	71 adolescent and adults aged 14 – 57 years (Community mean = 23.9, Clinical mean = 29)	ASD+ID community & clinical samples - ID = IQ<70 Community (52% mild/mod, 48% severe); clinical (78% mild/mod, 22% severe) - for results all ID levels grouped together	Community sample and clinical sample	Psychopathology in Autism Checklist (PAC) – developed for ASD population Clinical assessment	6 questions, mostly physiological: *fear in unusual situations, nervous, sweats, hyperventilates, shivering, distended pupils*	Physiological symptoms not easily identified by informants, PAC underreported diagnosis of Anxiety Anxiety was an issue for 37% of the community population
Hess, K. L. (2009)	313 children and adults aged 3-41 years (mean = 13)	ASD + ID & ASD alone - ID = IQ<70 (mean IQ = 55) - for results all ID levels grouped together	Community - specialist health services	Stress Survey Schedule (SSS) for Individuals with Autism and Other Pervasive Developmental Disabilities	62 items measuring stress across a number of subscales: *changes/threats, anticipation/uncertainty, unpleasant events, pleasant events, sensory/personal, food relation activity, social/environmental, ritual/related stress*	ASD+ID significantly higher stress scores for Changes/threats and Unpleasant events than ASD alone, but higher scores on most stress subscales across the board.

Joosten, A. V., Bundy, A. C., & Einfeld, S. L. (2009)	74 children aged 5-18 years (mean = 9.7)	ASD+ID & ID alone - ID = 2 standard deviations below the norm (mean IQ based on Vinelands score = 43.57) - for results all ID levels grouped together	Special schools	Developmental Behaviour Checklist (DBC) – developed for developmental delay population. Motivational Assessment Schedule Revised (MAS-R) – developed for a mainstream population	8 items on DBC – combination of physiological, separation anxiety/reaction to change. 4 items on MAS-R – capturing use of repetitive behaviour as a calming strategy, particularly in face of new or stressful experiences.	ASD+ID more likely to use RRBs when anxious, ID alone more likely to use RRBs for sensory seeking.
Joosten, A. V. & Bundy, A. C. (2010)	52 children aged 5-18 years (mean = 9.7)	ASD+ID & ID alone - ID = 2 standard deviations below the norm (mean IQ based on Vinelands score = 43.57) - for results all ID levels grouped together	Special schools	Developmental Behaviour Checklist (DBC) – developed for developmental delay population. Motivational Assessment Schedule Revised (MAS-R) – developed for a mainstream population	8 items on DBC – combination of physiological, separation anxiety/reaction to change. 4 items on MAS-R – capturing use of repetitive behaviour as a calming strategy, particularly in face of new or stressful experiences.	ASD+ID had a lower sensory threshold than ID alone, also have higher anxiety scores and increased repetitive behaviours to manage anxiety.
Joosten, A. V., Bundy, A. C., & Einfeld, S. L. (2012)	74 children aged 5-18 years (mean = 9.7)	ASD+ID & ID alone - ID = 2 standard deviations below the norm (mean IQ based on Vinelands score = 43.57) - for results all ID levels grouped together	Special schools	Developmental Behaviour Checklist (DBC) – developed for developmental delay population. Motivational Assessment Schedule Revised (MAS-R) – developed for a mainstream population	8 items on DBC – combination of physiological, separation anxiety/reaction to change. 4 items on MAS-R – capturing use of repetitive behaviour as a calming strategy, particularly in face of new or stressful experiences.	Individuals with ASD more likely to engage in repetitive behaviours motivate by anxiety during transition. Therefore, the same behaviour can have a different purpose in different contexts.
Moskowitz, L. J., Mulder, E., Walsh, C. E., McLaughlin, D. M., Zarcone, J. R., Proudfit, G. H., & Carr, E. G. (2013)	3 children ages: 6, 8 and 9 years old	All with ASD & Mild ID = based on adaptive behaviour scores: 56, 62, 70.	Local listserv of children with ASD	3 scales used: 1. Cues for Tension and Anxiety Survey Schedule. 2. Affex Facial Coding System for Negative Facial Expressions 3. Behavioural Relaxation Scale all developed for mainstream population	*Behavioural* (eg clinging, crying, repetitive questioning, plugging ears), *Affective* (appearance of anxiety on 4-point scale), *Physiological* (Heart Rate, RSA), *Problem Behaviour* (eg. Yelling, absconding, tantrums)	Fewer problem behaviours seen in low-anxiety contexts. High anxiety related to high HR, low RSA and high problem behaviours.

Moskowitz, L. J., Walsh, C. E., Mulder, E., McLaughlin, E., Hajcak, G., Carr, E. G. & Zarcone, J. R. (2017)	3 children ages: 6, 8 and 9 years old	All with ASD & Mild ID = based on adaptive behaviour scores: 56, 62, 70.	Local listserv of children with ASD	3 scales used: 1. Cues for Tension and Anxiety Survey Schedule. 2. Affex Facial Coding System for Negative Facial Expressions 3. Behavioural Relaxation Scale all developed for mainstream population	*Behavioural* (eg clinging, crying, repetitive questioning, plugging ears), *Affective* (appearance of anxiety on 4-point scale), *Physiological* (Heart Rate, RSA), *Problem Behaviour* (eg. Yelling, absconding, tantrums)	With multi-modal treatment program (Positive Behaviour Support and CBT), anxiety scores reduced, problem behaviours observed reduced. Also showed a reduction in heart rate and an increase in RSA – showing better coping and tolerance in previously high stress situations.
Sukhodolsky, D. G., Scahill, L., Gadow, K. D., Arnold, L. E., Aman, M. G., McDougle, C. J., . . . Vitiello, B. (2008)	171 children aged 5-14 years (mean = 8.2)	ASD + ID & ASD alone - ID = IQ<70 (mild=23%, mod=14%, severe=15%, profound=10%) - for results all ID levels grouped together	Clinical trials of children with ASD enrolled in medication trials through RUPP Autism Network	Child and Adolescent Symptom Inventory (CASI) - developed for mainstream population	20 anxiety specific questions - mixture of physiological symptoms and cognitive markers; based on DSM-IV anxiety disorder criteria: GAD, Simple Phobia, Panic disorder, Somatic Disorder, Social Phobia, Separation Anxiety	Higher IQ, functional language and stereotyped behaviours associated with higher anxiety, also associated with Social Phobia Lower IQ associated with hyperactivity and Inappropriate speech

Evaluation of Selected Papers

The two-step evaluation process, adapted from MacNeil et al. (2009), included looking at how anxiety was identified and then the methodology of research conducted. The results of the first evaluation process, focusing on how anxiety was identified, are shown in Table 3. Eight of the nine papers were evaluated, with a final score range of 1 to 4 and an average score of 2.75. Higher scores indicate that more elements for a multimodal diagnostic process were employed. The ninth paper, Helverschou et al. (2008), was not evaluated, as it was the only paper that did not directly diagnose anxiety, but instead outlined the symptoms clinicians assigned to psychiatric conditions. The results of the methodology evaluation are available in Table 4. Helverschou et al. (2008) was included, although the different nature of the paper meant that it was difficult to accurately score. Of the nine papers, the range of final scores was 2 to 6 with an average of 4 out of 8, with higher scores indicating a more comprehensive methodology.

Table 2

Evaluation of assessment methodology: using recommendations from literature

Researchers	Is a clinical interview conducted?	Are Anxiety Rating Scales used?	Is direct observation used to assess anxiety?	Are physiological assessment measures used?	Are multiple informants used to assess anxiety?	Is comparative data available for ASD populations for any of the anxiety instruments?	Total score (number of yes)
Helverschou, Bakken & Martinsen (2008)	N/A	N/A	N/A	N/A	N/A	N/A	N/A
Helverschou & Martinsen (2011)	YES	YES - PAC	NO	NO - only as part of rating scale	YES - carers	NO - but designed for disability population	3
Hess (2009)	NO	YES - SSS	NO	NO	NO	NO - but designed for disability population	1
Joosten, Bundy & Einfeld (2009)	NO	YES - DBC	YES	NO	YES - teacher + author	NO - but designed for disability population	3
Joosten & Bundy (2010)	NO	YES - DBC	YES	NO	YES - teacher + author	NO - but designed for disability population	3
Joosten, Bundy & Einfeld (2012)	NO	YES - DBC	YES	NO	YES - teacher + author	NO - but designed for disability population	3
Moskowitz et al. (2013)	NO	YES – ind.	YES	YES	YES - 2 blind raters	NO	4
Moskowitz et al. (2017)	NO	YES – ind.	YES	YES	YES - 2 blind raters	NO	4
Sukhodolsky et al. (2008)	NO	YES - CASI	NO	NO - only as part of rating scale	NO	NO	1

Table 3

Methodological quality rating system

Researchers	Is the sample well defined?	Does the current author confirm the ASD diagnosis?	Is there a current comparison group?	Is the comparison group matched?	Are the data collection methods standardised?	Are the anxiety instruments reliable?	Are the anxiety instruments valid?	Are the interpretations appropriate?	Total validity score
Helverschou, Bakken & Martinsen (2008)	YES	NO	NO	NO	NO	Not stated	Not stated	YES	2
Helverschou & Martinsen (2011)	YES	NO	YES	NO	YES	YES	YES	YES	6
Hess (2009)	NO	NO	NO	NO	YES	YES	YES	YES	4
Joosten, Bundy & Einfeld (2009)	YES	NO	YES	NO	YES	Not stated	Not stated	YES	4
Joosten & Bundy (2010)	YES	NO	YES	NO	YES	Not stated	Not stated	YES	4
Joosten, Bundy & Einfeld (2012)	YES	NO	YES	NO	YES	Not stated	Not stated	YES	4
Moskowitz et al. (2013)	YES	NO	NO	NO	YES	Not stated	Not stated	YES	3
Moskowitz et al. (2017)	YES	NO	NO	NO	YES	Not stated	Not stated	YES	3
Sukhodolsky et al. (2008)	NO	YES	YES	NO	YES	YES	YES	YES	6

Anxiety in ASD and ID

Four studies were identified that focused on the way anxiety was identified for individuals with ASD and ID. Moskowitz et al. (2013) and Moskowitz, Walsh et al. (2017) looked at three case studies of children with ASD and ID and the way anxiety could be identified through a multimodal assessment process. The authors found that when faced with highly stressful situations (compared to low stress situations), children with ASD and ID showed high heart rate, low respiratory sinus arrhythmia (both measured through a portable electrocardiogram) and high levels of problem behaviours (such as screaming/yelling, elopement, hitting others and tantrum type behaviour). This physiological pattern is identical to that expected of non-ASD individuals experiencing anxiety, with the exception of the increase in problem behaviours (Moskowitz et al., 2013).

Further research by Moskowitz, Walsh et al. (2017) focused on the reduction of anxiety with treatment. They found that as anxiety markers decreased, problem behaviours and heart rate also decreased, while respiratory sinus arrhythmia was conversely increased, indicating that the children were more able to regulate their stress, displaying better coping and tolerance.

Helverschou et al. (2008) postulated that psychiatric conditions could be separated from ASD and therefore identified through a unique set of symptoms for individuals with ASD and ID. Through a panel of 9 clinicians, 254 symptoms were rated according to which condition they best described for individuals with ASD and ID. Of the 17 symptoms found to identify anxiety, 12 were physiological, such as *hyperventilates*, *changes in muscle tone* and *sweats*; while only five were considered to be more cognitive or behavioural such as *fear in specific situations* and *increased fear and avoidance*. The researchers note that the more cognitive symptoms generally associated with anxiety, such as *excessive worry* and *fear of specific situations*, were less useful in clinical settings for diagnosing anxiety in this population, finding that the physiological symptoms were more applicable and reliable.

This work led to the development of an ASD specific psychiatric measurement tool, which was tested on community and clinical samples of individuals with ASD and ID by Helverschou and Martinsen in 2011. Caregivers completed the Psychopathology in Autism Checklist (PAC) (Helverschou et al., 2009), with those in the clinical sample also receiving a clinical assessment. The authors noted that the anxiety subscale on the PAC was not a good predictor of anxiety diagnosis when compared to a clinical assessment, with carers consistently underreporting or failing to identify physiological symptoms of anxiety. However, all those with anxiety had high scores on the PAC general adjustment issues subscale.

Anxiety in ASD and ID compared to ID alone. Three studies based on the same data considered the way anxiety may manifest differently in individuals with ASD and ID compared to those with ID alone: Joosten et al. (2009, 2012) and Joosten and Bundy (2010). In their 2009 paper, they reported that children with ASD and ID were more likely to use repetitive behaviours as a result of anxiety than for sensory seeking purposes, while the reverse was true for children with ID alone. Joosten and Bundy (2010) included sensory processing with their existing anxiety data, finding that the children with ASD and ID were more sensitive to and avoidant of sensory experiences than those with ID alone. While in 2012, Joosten et al. looked more specifically at the motivation to perform repetitive behaviours in different situations. They found that for those with ASD and ID, transitions between tasks were particularly challenging and they were therefore more likely to exhibit repetitive behaviours motivated by escape and anxiety than those with ID alone.

Sukhodolsky et al. (2008) took the information gathered for a medication trial to see how parents rated anxiety symptoms in children with ASD with and without ID. They found that those with ASD alone showed higher IQ and functional language levels, as well as higher stereotyped behaviours and higher anxiety levels. In this group, higher IQ was also associated with higher social impairments. Those with a dual diagnosis of ASD and ID had higher levels of hyperactivity and inappropriate speech.

Hess (2009), in her doctoral thesis discussed anxiety as an outcome of increased stress and considered how individuals with ASD (with and without an ID) respond to stress across an array of settings and scenarios, using the Stress Survey Schedule (SSS). This research found that those with both ASD and ID were noted to have higher stress levels than those with ASD alone on six out of the eight stress subscales. While anxiety is discussed as an outcome of increased stress, a more specific anxiety scale or diagnostic format was not used. As such the impact of the stressors and the potential relationship to clinically significant levels of anxiety is unknown.

Anxiety Measurement Tools and Diagnostic Methods. Different diagnostic methods and anxiety measurement tools were used in each study. Three studies employed direct observation of the participants, one study incorporated a clinical interview and one study included direct measurement of physiological data. Of particular note is the lack of consistency in the anxiety measurement tools utilised, five different rating scales were used between the five research groups. None of these measures had normative data available for an ASD population, though three were developed for a disability population (details in Table 2). In looking at the items on these anxiety scales, there is a lack of consistency in the identifying markers of anxiety, with only a single item appearing on each of the scales: *fear of a specific situation or object* (or similar). Table 5 presents an overview of the five measurement tools and indicates whether anxiety markers were present across the subcategories identified. None of the tools covered all of the five subcategories, with a range of two to four categories being addressed through individual anxiety markers.

Table 4

Subcategories covered by individual items assessing anxiety on measurement tools

	CASI	DBC P	MAS-R	PAC	SSS
Physiological - Anxious Anxiety	X	X	X	X	
Cognitive - Separation Anxiety	X	X			X
Cognitive - Performance Anxiety / Language Based	X				X
Cognitive - Uncertainty with situation / location / expectation	X	X	X	X	X
Behaviour Observations		X			X

When looking at these measurement tools in more detail, it is clear that there is only limited overlap in the items used to assess or the method used to identify anxiety. Both the Psychopathology in Autism Checklist (PAC; Helverschou et al., 2009) and the Child and Adolescent Symptom Inventory (CASI; Gadow & Sprafkin, 1994 and 1997) include a combination of both physiological and cognitive symptoms. The PAC relies more heavily on the physiological with only two cognitive items, while the majority of the CASI items have a more cognitive focus. The Developmental Behaviour Checklist (DBC; Einfeld & Tonge, 1995) relies on eight diverse symptoms to capture anxiety, mostly behavioural in nature, including such items as *easily distressed when alone, shy, fussy eater* and *distress when small changes occur*. In contrast, the Stress Survey Schedule (SSS; Groden et al., 2001) and the Motivation Assessment Schedule-Revised (MAS-R; Joosten et al., 2009) focus on situational stress and anxiety, and include lists of situations that may induce stress or cause behaviours to occur more frequently, rather than a list of symptoms.

Discussion

The question of the way anxiety is identified and diagnosed in individuals with ASD, and ID is not easily answered. No clear pattern emerges in the papers reviewed, though it does appear to be the case that anxiety presents differently for those with ASD and ID than for those with either ASD or ID alone. Several key considerations have emerged from this review, particularly around the way anxiety is measured and diagnosed and the impact that this has on research findings and the conclusions drawn.

Anxiety Symptoms

Previous research has found a considerable overlap in the symptoms of ASD and anxiety, leading to a lack of clarity around how to distinguish between the two conditions (Kerns et al., 2014). To address this, Helverschou et al. (2008) set out to find symptoms of anxiety that were completely separate to ASD. However, Joosten et al. (2009 and 2012) found that repetitive behaviours, a key component in ASD diagnosis, were actually exacerbated by elevated anxiety. This relationship has also been found by Rodgers et al. (2012) and Spiker et al. (2011), with Kerns et al. (2014) suggesting that the presence of a co-occurring disorder is likely to be exhibited through ASD symptom amplification rather than a separate set of behaviours. Therefore, it would appear unwise to ignore ASD characteristics in favour of a unique set of symptoms to identify anxiety, as this could result in under reporting diagnosis and therefore treatment.

Joosten and Bundy (2010) also found that those with ASD and ID were more likely to be sensitive to and avoidant of sensory experiences than those with ID alone, while also showing elevated anxiety rates. It may be that sensory profiles offer another possibility for identifying anxiety in those with ASD and ID.

Moskowitz et al. (2013) and Moskowitz, Walsh et al. (2017) found that while individuals with ASD and ID also showed the same physiological patterns of stress when anxious as non-ASD peers, the presence of increased problem behaviours was a key indicator of the presence of anxiety in individuals with ASD and ID. A measure of problem behaviour was not included in this study, but rather individual profiles of problem behaviour were developed for each case study and frequency used for measurement. Helverschou and Martinsen (2011) likewise found that general adjustment difficulties were related to elevated anxiety scores on the PAC. Likewise, Rzepecka et al. (2011) have found that in children with ASD and ID, challenging behaviours (such as physically injuring themselves or others) were associated with an increase in anxiety symptoms. These behaviours may be more identifiable by carers than physiological markers, which were routinely under reported.

The direct measurement of physiological symptoms of anxiety has several limitations for the ASD and ID population. Moskowitz et al. (2013 and Moskowitz, Walsh et al., 2017) found that in order for the participants to tolerate the electrodes for the electrocardiogram to be administered, extensive prior training was required. This would be a considerable limitation given the extreme variability in the sensory profile of individuals with ASD and would necessarily exclude those who could not tolerate the process, as the process itself could also provoke increased stress and anxiety. The external measurement of physiological symptoms of anxiety is unlikely to represent a practical means of assessing anxiety in clinical settings.

Sukhodolsky et al. (2008) found that there was a relationship between anxiety measures and ASD symptomology, though this was particularly true for those with higher IQs. They found that higher anxiety co-occurred with higher social impairments for those with higher IQs. This research would therefore suggest that children with ASD alone experience more anxiety than those with ASD and ID. While this does not help elucidate the situation for those with ASD, ID and anxiety, it does suggest that there is overlap between the symptoms of ASD and anxiety and that trying to completely separate the two may not be useful.

Anxiety Measurement Tools

Of the papers reviewed, five different anxiety measures were used, with little overlap between these measures in terms of the anxiety domains they addressed. Three of the measurement tools (DBC, PAC and SSS) were created specifically for a disability population but were not standardised specifically for an ASD and ID population. The CASI and MAS-R were created for mainstream populations and do not have normative data for ASD or ASD and ID populations.

With such a variety of symptoms being used to diagnose anxiety, it is hardly surprising that results are divergent. Sukhodolsky et al. (2008) used the CASI with items that are largely cognitive in nature and not normed for an ASD or ASD and ID population. It is therefore not surprising that they found that those with a higher IQ experienced higher anxiety. Helverschou et al. (2008) mention that cognitive symptoms were not useful in clinical settings when considering diagnosis for those with ASD and ID. Therefore, measurement tools that focus on cognitively based anxiety symptoms are likely to underestimate the anxiety experienced by those with ASD and ID. This may also explain the differences in research findings between those with and without ID, the focus of the anxiety measurement tool used may affect the outcomes. The lack of consistency between the measures used to assess anxiety makes it challenging to compare studies and draw conclusions.

This review found that there is currently no anxiety measure being used universally for research that is standardised for an ASD and ID population. This is a key issue in the conclusions being drawn from research into the diagnosis of anxiety in this population, with MacNeil et al. (2009) identifying use of an ASD appropriate tool as essential to reliable diagnosis. Gronduis and Aman (2012) similarly found that there was no single outstanding measure with sound internal structure, reliable data and that was validated for use with an ASD population, let alone an ASD and ID cohort. Attempts have been made to develop an ASD specific anxiety measure, such as the Anxiety Scale for Children with Autism Spectrum Disorder (ASC-ASD; Rodgers, Wigham et al., 2016) and the Anxiety Disorders Interview Schedule-Child/Parent: Parent version (Silverman & Albano, 1996), Autism Spectrum Addendum (ADIS/ASA) (Kerns et al., 2014). These measures have started to be used in research with some promising results (den Houting et al., 2018; Kerns et al., 2017), however have not been standardised for use with an ASD and ID population, nor has such a measure been developed for an adult population.

Diagnostic Difficulties

This research found that none of the papers reviewed used multimodal diagnostic techniques in identifying anxiety within individuals with ASD and ID, with most relying heavily on questionnaire-based information. This is despite research suggesting that accurate diagnosis requires a more rigorous testing method, such as that posited by MacNeil et al. (2009). The lack of a consistent diagnostic procedure may also be impacting our ability to accurately identify and diagnose anxiety in this complex population.

The authors of the included studies consistently discussed the difficulties with diagnosis for individuals with both communication and cognitive limitations. Of particular note is the need to rely heavily on the information gathered from family and carers, where identification and attribution of behaviours can alter the information gathered. Helverschou and Martinsen (2011) in particular noted the need for a multimodal diagnostic process, as relying on the identification of both cognitive and physiological symptoms by caregivers would likely under report the true incidence rate. Bailey and

Andrews (2003) likewise note that relying solely on carer feedback can be difficult as a carer’s experience of life may differ from that of the individual with ID for whom they care. This difference in experience can make it problematic to attribute cause to behaviour or to identify the difference between pathological and adaptive behaviours. This is a considerable difficulty given how much carer feedback is relied upon for the diagnostic process within both research and clinical settings.

Identifying and diagnosing anxiety for individuals with ASD and ID is compounded by the considerable overlap between ASD and anxiety symptoms, and by the communication difficulties inherent to those with both ASD and ID. The cognitive based symptoms of excessive worry and fear of specific situations are less useful clinically for diagnosis, and the physiological symptoms, while in theory more observable, are often not identified by carers when feedback is sort. It may be that considering more observable behaviours patterns and changes over time offers a more reliable means of identifying anxiety in those with ASD and ID.

Limitations

This study has provided an overview of the current research in this area, however limited research was available to allow for full elucidation into how anxiety can be identified in individuals with ASD and ID. In fact, only nine studies from the last 24 years were found to meet the selection criteria, clearly pointing to a need for further research in this area. This small catchment may have been affected by the search terms chosen. While Asperger’s Syndrome was deliberately excluded, Pervasive Developmental Disorder - Not Otherwise Specified was also not utilised. Theoretically there is little overlap between these conditions and ID, however this may not have been the practice in research and therefore by excluding, some useful research may have been missed.

The research papers included here for review grouped all levels of ID together, which given the research finding variability across the ID range, significantly affects the generalisability of the results. There is compelling research that points to differential presentation across the ID range, and therefore by grouping all ID groups together, we could be missing valuable differences that occur between mild and severe ID populations. While a large age range was represented in the papers reviewed, there was no mention of how anxiety may change across an individual's lifespan. Davis et al. (2011) found that anxiety symptoms alter in presentation and intensity across the lifespan, particularly noting that anxiety symptoms may change with age. This would therefore require researchers and clinicians to consider both cognitive and developmental stages when considering how to identify anxiety accurately across the lifespan of an individual with ASD.

This study provides a preliminary overview of current research within this area, but there may be a subgroup within the ASD and ID population that have not been identified given the difficulties with diagnosis and definitions identified.

Future Research

Future research should further explore the correlates and behavioural patterns of anxiety for those with ASD and ID, as this may result in more accurate and useful diagnostic processes. Of particular interest should be consideration of how anxiety presents differently across the ID range, as it seems likely this will vary considerably between mild and severe levels of ID. Also, of interest is how observable behaviours (such as difficult, challenging or problem behaviours), may offer a more reliable way of identifying anxiety in the group that relies so heavily on carer feedback.

The development of an anxiety measure that takes into account the unique behaviours and changes in presentation over time of those with ASD and ID, could also result in more reliable identification and diagnosis. Without this, individuals cannot receive the treatment and support that may significantly impact their quality of life.

Conclusion

There is a lack of consensus regarding the way anxiety can be identified in individuals with ASD and ID, though it does appear that a different pattern from ASD alone does exist, with suggestions of other behavioural markers to identify anxiety being a useful consideration. The lack of specifically standardised measurement tools is an issue, as the wide variety of tools available seem to have limited consistency between them, despite claiming to measure the same phenomena. There has also been an emphasis on creating a separate symptom criterion for the diagnosis of anxiety as distinct from ASD, which may actually be impacting our ability to identify and diagnosis accurately.

References

American Psychological Association (APA) (2013). *Diagnostic and statistical manual of mental disorders (5th ed.)* Arlington, VA: American Psychiatric Publishing.

Bailey, N. M. and T. M. Andrews (2003). "Diagnostic Criteria for Psychiatric Disorders for use with Adults with Learning Disability/Mental Retardation (DC-LD) and the diagnosis of anxiety disorders: a review." *Journal of Intellectual Disability Research, 47,* 50-61. doi: 10.1046/j.1365-2788.47.s1.25.x

Bradley, E. A., Summers, J. A., Wood, H. L., & Bryson, S. E. (2004). Comparing rates of psychiatric and behavior disorders in adolescents and young adults with severe intellectual disability with and without autism. *Journal of Autism and Developmental Disorders, 34*, 151 - 161. doi: 0162-3257/04/0400-0151/0

Brereton, A. V., Tonge, B. J., & Einfeld, S., L. (2006). Psychopathology in children and adolescents with autism compared to young people with intellectual disability. *Journal of Autism and Developmental Disorders, 36*, 863 - 870. doi: 10.1007/s/10803-006-0125-y

Colquhoun, H. L., Levac, D., O-Brien, K. K., Straus, S., Tricco, A. C., Perrier, L., Kastner, M., Moher, D. (2014). Scoping reviews: time for clarity in definition, methods and reporting. *Journal of Clinical Epidemiology, 67*, 1291-1294. doi: 10.1016/j.jclinepi.2014.03.013.

Davis, T. E., et al. (2011). "Anxiety symptoms across the lifespan in people diagnosed with Autistic Disorder." *Research in Autism Spectrum Disorders, 5,* 112 - 118. doi: 10.1016/j.rasd.2010.02.006

Deb, S., & Prasad, K. (1994). The prevalence of autistic disorder among children with a learning disability. *British Journal of Psychiatry, 165*, 395-399. doi: 10.1192/bjp.165.3.395

den Houting, J., Adams, D., Roberts, J., Keen, D. (2018). Exploring anxiety symptomatology in school-aged autistic children using an autism-specific assessment. *Research in Autsm Spectrum Disorders, 50*, 73-82. doi: 10.1016/j.rasd.2018.03.005

Einfeld, S. L., & Tonge, B. J. (1995). The Developmental Behaviour Checklist: the development and validation of an instrument to assess behavioural and emotional disturbance in children and adolescents with mental retardation. *Journal of Autism and Developmental Disabilities, 25*, 81-104. doi: 10.1007/BF02178498

Einfeld, S. L. &B. J. Tonge (1996). "Population prevalence of psychopathology in children and adolescents with intellectual disability: II epidemiological findings." *Journal of Intellectual Disability Research, 40,* 99-109. doi: 10.1046/j.1365-2788.1996.768768.x

Gadow, K. D. & Sprafkin, J. (1994). *Child symptom inventory-4.* Stony Brook, NY: Checkmate Plus.

Gadow, K. D. & Sprafkin, J. (1997). *Adolescent symptom inventory-4 screening manual*. Stony Brook, NY: Checkmate Plus.

Groden, J., Diller, A., Bauseman, M., Velicer, W., Norman, G. and Cautela, J. (2001). The development of a stress survey schedule for persons with autism and other developmental disabilities. *Journal of Autism and Developmental Disabilities*, *31*. 207-217. doi: 10.1023/A:101075530

Groden, J., Baron, M. G., & Groden, G. (2006). Assessment and Coping Strategies. In M. G. Baron, J. Groden, G. Grode, & L. P. Lipsitt (Eds.), *Stress and Coping in Autism* (pp. 15 - 41). New York: Oxford University Press.

Grondhuis, S. N., & Aman, M. G. (2012). Assessment of anxiety in children and adolescents with autism spectrum disorders. *Research in Autism Spectrum Disorders 6*, 1345–1365. doi: 10.1016/j.rasd.2012.04.006

Helverschou, S. B., Bakken, T. L., & Martinsen, H. (2008). Indentifying symptoms of psychiatric disorders in people with autism and intellectual disability: An empirical conceptual analysis. *Mental Health Aspects of Developmental Disabilities*, *11*, 105-115.

Helverschou, S. B., Bakken, T. L., & Martinsen, H. (2009). The psychopathology in autism checklist (PAC): a pilot study. *Research in Autism Spectrum Disorders, 3*, 179-195. doi: 10.1016/j.rasd.2008.05.004

Helverschou, S. B., & Martinsen, H. (2011). Anxiety in people diagnosed with autism and intellectual disability: Recognition and phenomenology. *Research in Autism Spectrum Disorders, 5*, 377 - 387. doi: 10.1016/j.rasd.2010.05.003

Hermans, H., van der Pas, F. & Evenhuis, H. (2011). "Instruments assessing anxiety in adults with intellectual disabilities: A systematic review." *Research in Developmental Disabilities, 32(3),* 861-870. doi: 10.1016/j.ridd.2011.01.034

Hess, K. L. (2009) *Stress for individuals with autism spectrum disorders: effects of afe, gender and intelligence quotient.* PhD Thesis: Georgia State University, Ann Arbor.

Hill, J., & Furniss, F. (2006). Patterns of emotional and behavioural disturbance associated with autistic traits in young people with severe intellectual disabilities and challenging behaviours *Research in Developmental Disabilities, 27*, 517–528. doi: 10.1016/j.ridd.2005.07.001

Joosten, A. V., Bundy, A. C., & Einfeld, S. L. (2009). Intrinsic and extrinsic motivation for stereotypic and repetitive behaviour. *Journal of Autism and Developmental Disorders, 39*, 521 - 531. doi: 10.1007/s10803-008-0654-7

Joosten, A.V. & Bundy, A.C. (2010). Sensory processing and stereotypical and repetitive behaviour in children with autism and intellectual disability. *Australian Occupational Therapy Journal, 57(6),* 366 - 372. doi: 10.1111/j.1440-1630.2009.00835.x

Joosten, A. V., Bundy, A. C., & Einfeld, S. L. (2012). Context influences the motivation for thereotypic and repetitive behaviour in children diagnosed with intellectual disability with and without autism. *Journal of Applied Research in Intellectual Disabilities*, *25*, 262-270. doi: 10.1111/j.1468-3148.2011.00663.x

Kanner, L. (1943). Autistic disturbances of affective contact. *Nervous Child, 2*, 217-250.

Kerns, C. M., Kendall, P. C., Berry, L., Souders, M. C., Franklin, M. E., Schultz, R. T., Miller, J., Herrington, J. (2014). Traditional and atypical presentations of anxiety in youth with autism spectrum disorder. *Journal of Autism and Developmental Disoders, 44*, 2851-2861. doi: 10.1007/s10803-014-2141-7

Kerns, C. M., et al. (2017). "Anxiety Disorders Interview Schedule-Autism Addendum: Reliability and Validity in Children with Autism Spectrum Disorder." *Journal of Clinical Child and Adolescent Psychology, 46(1)*, 88-100. doi:10.1080/15374416.2016.1233501

Lainhart, J. E., & Folstein, S. E. (1994). Affective disorders in people with autism: a review of published cases. *Journal of Autism and Developmental Disorders, 24*, 587 - 601.

Lecavalier, L. (2006). Behavioral and emotional problems in young people with pervasive developmental disorders: relative prevalence, effects of subject characteristics and empirical classification. *Journal of Autism and Developmental Disorders, 36*(8), 1101-1114. doi: 10.1007/s10803-006-0147-5

MacNeil, B. M., Lopes, V. A., & Minnes, P. M. (2009). Anxiety in children and adolescents with Autism Spectrum Disorders. *Research in Autism Spectrum Disorders, 3*, 1 - 21. doi: 10.1016/j.rasd.2008.06.001

Militerni, R., Bravaccio, C., Falco, C., Fico, C., & Palermo, M. T. (2002). Repetitive behaviours in autistic disorder. *European Child and Adolescent Psychiatry, 11*, 210 - 218. doi: 10.1007/s00787-002-0279-x

Morgan, K. (2006). Is Autism a stress disorder? What studies of nonautistic populations can tell us. In M. G. Baron, J. Groden, G. Groden, & L. P. Lipsitt (Eds.), *Stress and Coping in Autism* (pp. 129 - 182). New York, NY: Oxford University Press.

Moskowitz, L. J., Mulder, E., Walsh, C. E., Mclaughlin, D. M., Zarcone, J. R., Proudfit, G. H. & Carr, E. G. (2013). A multimethod assessment of anxiety and problem behaviour in children with autism spectrum disorders ad intellectual disability. *American Journal on Intellectual and Developmental Disabilities, 118*, 419-434. doi: 10.1352/1944.7558.118.6.419

Moskowitz, L. J., Walsh, C. E., Mulder, E., McLaughlin, E., Hajcak, G., Carr, E. G. & Zarcone, J. R. (2017). Intervention for anxiety and problem behaviour in children with autism spectrum disorder and intellectual disability. *Journal of Autism and Developmental Disorders, 47*, 3930-3948. doi: 10.1007/s10803-017-3070-z

Murphy, O., Healy, O., & Leader, G. (2009). Risk factors for challenging behaviors among 157 children with autism spectrum disorder in Ireland. *Research in Autism Spectrum Disorders, 3*, 474 – 482. doi: 10.1016/j.rasd.2008.09.008

Pyles, D., Muniz, K., Cade, A., & Silva, R. (1997). A behavioural diagnostic paradigm for integrating behaviour-analytic and psychpharmacological interventions for people with a dual diagnosis. *Research in Develomental Disabilities, 18*, 185-214. doi:10.1016/S0891-4222(97)00003-6

Rivard, M., Terroux, A., Mercier, C., & Parent-Boursier, C. (2015). Indicators of intellectual disabilities in young children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 45*, 127-137. doi: 10.1007/s10803-014-2198-3

Rodgers, J., Glod, M., Connolly, B., & McConachie, H. (2012). The Relationship Between Anxiety and Repetitive Behaviours in Autism Spectrum Disorder. *Journal of Autism and Developmental Disorders, 42*, 2404-2409. doi: 10.1007/s10803-012-1531-y

Silverman, W. K. and A. M. Albano (1996). *The Anxiety Disorders Inte4rview Schedule for Children (ADIS-C/P).* San Antonio, TX, Psychological Corporation.

Simonoff, E., A., P., Charman, T., Chandler, S., Loucas, T., & Baird, G. (2008). Psychiatric disorders in children with autism spectrum disorders: prevalence, comorbidity, and associated factors in a population-derived sample. *Journal of the American Academy of Child and Adolescent Psychiatry, 47*, 921-929. doi: 10.1097/CHI.0b013e318179964f

Spiker, M. A., Lin, C. E., Van Dyke, M., & Wood, J. (2011). Restricted interests and anxiety in children with autism. *Autism, 16*, 306-320. doi: 10.1177/1362361311401763

Sukhodolsky, D. G., Scahill, L., Gadow, K. D., Arnold, L. E., Aman, M. G., McDougle, C. J., . . . Vitiello, B. (2008). Parent-Rated Anxiety Symptoms in Children with Pervasive Developmental Disorders: Frequency and Association with Core

Autism Symptoms and Cognitive Functioning. *Journal of Abnormal Child Psychology, 36*, 117-128. doi: 10.1007/s10802-007-9165-9

Tantam, D. (2000). Psychological disorder in adolescents and adults with Asperger syndrome. *Autism, 4*, 47-62.

As this scoping review was published in 2019, the search was conducted again through PsychInfo, Proquest, and Medline to capture publications from 2019 to March 2022, following the same search terms and parameters. As a result, 330 possible papers were found; however, no additional papers were found that met the search question, that is, addressing the way anxiety was identified and diagnosed in individuals with ASD and ID.

Challenges With Identifying and Diagnosing Anxiety in ASD and ID

The above paper outlines specific research that addresses how anxiety can be identified in those with comorbid ASD and ID. Difficulties with the identification process are considerable, with issues including consensus as to what measurement tools are actually measuring, symptom amplification, and diagnostic overshadowing all being issues. Given the paucity of research into how anxiety affects those with ASD and ID, the key themes found in the scoping review will now be elaborated on, with additional research included. This research includes studies into anxiety and ASD or ID alone, but also includes those studies that, due to the focus or nature of the research, were not included in the scoping review.

Communication and Information Processing

Prizant (1996) reported that up to 50% of individuals with autism/pervasive developmental delay will not use speech as their primary form of communication. This limits their ability to label their internal experience and communicate their thoughts and feelings to others (Gjevik et al., 2011; Helverschou & Martinsen, 2011). Lainhart and

Folstein (1994) also pointed out individuals with ASD can find abstract thinking challenging, which can affect their understanding of internal concepts and therefore impact their ability to describe their mood and emotions. Given that psychiatric diagnosis is based on client interview and information gathering, this is a considerable limitation in the availability of accurate data on which a professional can base a decision and, therefore, diagnosis (Bradley et al., 2004; Grondhuis & Aman, 2012).

The presence of a co-occurring ID further complicates the diagnostic process. The deficits in cognitive functioning can lead to further difficulties in understanding and expressing more complex emotions and processes, such as those underpinning psychiatric conditions (Witwer & Lecavalier, 2010). Grondhuis and Aman (2012) pointed out that individuals with ASD and ID can lack insight into their own experience, making it difficult for them to describe their own symptoms. Communication difficulties can limit parent and carer awareness of internal symptoms and emotions, increasing the likelihood that a comorbid diagnosis will be overlooked.

Symptom Amplification

Research into comorbid diagnosis has yet to find a unique criterion that would allow for clear and consistent differentiation to occur between ASD and a comorbid psychiatric disorder (Helverschou et al., 2009). This can be difficult when looking at the comorbidity with ASD, where the presence of a co-occurring disorder may be exhibited through ASD symptom amplification rather than a separate set of behaviours (Kerns et al., 2014). Lainhart and Folstein (1994) stated that depression may be most evident not through mood changes, but through increased self-injury and ASD symptom severity, such as echolalia and RRBs. Joyce et al. (2017), Spiker et al. (2011), and Rodgers et al. (2012) all reported that for individuals with ASD, anxiety appears to exacerbate RRBs. A possible motivation for RRBs has been suggested to be an attempt at managing anxiety and coping with additional stress (Davis et al., 2011; Joosten et al., 2009);

therefore, seeing increased levels of RRBs as a result of anxiety would not be unexpected.

This issue is referred to as baseline exaggeration, whereby behavioural changes indicate that the emergence of a psychiatric disorder is an exacerbation or intensifying of already existing behaviours. It is therefore easy to miss as it can be read as an increase in ASD symptom severity rather than as the development of an additional condition. Traditional definitions of comorbid diagnosis seek to find distinctly separate behavioural markers for separate disorders. In the case of baseline exaggeration in ASD, this would result in diminished diagnoses of comorbid disorders (Leyfer et al., 2006).

With baseline exaggeration in mind, it is therefore important to consider behaviour changes over time, rather than concentrating on a single assessment point when diagnosing anxiety. Lainhart (1999) considered it critical to evaluate the way behaviours change from the individual's own baseline, rather than relying on a screening tool with a single point of information. Matson and Nebel-Schwalm (2007) also pointed out the need to have periodic monitoring, in order to differentiate between the evolving psychopathology and the wax and wane of ASD symptoms over time as a function of development.

Diagnostic Overshadowing and Differential Diagnosis

The similarities between characteristics of ASD and anxiety can lead to diagnostic overshadowing, that is, the attribution of behaviours or symptoms to the disability diagnosis (ASD) rather than to a comorbid mental health concern (Helverschou & Martinsen, 2011). In this way, the ASD diagnosis overshadows the presence of a secondary diagnosis. For example, obsessions and panic attacks may be labelled as behaviour problems synonymous with ASD rather than as symptoms of a comorbid anxiety disorder (Tsai, 2006).

Challenging behaviours such as self-injurious behaviour, aggression, and property damage are often attributed to ASD, without consideration of a possible comorbid diagnosis. Challenging behaviours are more likely to present in individuals with ASD and ID (Murphy et al., 2009) and are associated with the severity of ID (McCarthy et al., 2010). Challenging or problem behaviour is an observable trait and one that has been consistently predictive of heightened anxiety in those with ASD with and without ID (Bitsika & Sharpley, 2016; Moskowitz et al., 2013; Rzepecka et al., 2011). Challenging behaviours are consistently reported by carers of individuals with ASD; Murphy et al. (2009) found that among a group of individuals with ASD and ID, 82% displayed challenging behaviours (including self-injury, aggression, and stereotyped behaviours), with 96% showing more than one form. Nicholls et al. (2020), in a review of incidence rates of challenging behaviours within special schools in the UK, found that 53% of the children with ID displayed challenging or problem behaviours, with co-occurring ASD being the most significant and common predictive variable. Problem or challenging behaviours therefore represent a significant issue for individuals with ASD and ID and for those who support them. Rzepecka et al. (2011) have found that in children with ASD and ID, challenging behaviours were associated with an increase in anxiety symptoms. They postulated that challenging behaviours could be considered an atypical manifestation of psychiatric disorder, or behavioural equivalents, rather than a symptom or behaviour of ASD itself. Moskowitz et al. (2013) and Moskowitz, Walsh et al. (2017) likewise considered challenging behaviour as an indicator of additional difficulties, rather than attributing it to an ASD diagnosis, as detailed in the scoping review above.

Another diagnostic issue for consideration is that of differential diagnosis. For example, there is considerable overlap of behaviours between ASD and psychiatric disorders such as schizophrenia, anxiety, depression, and OCD (Bakken et al., 2010;

Helverschou et al., 2009). A single behaviour or symptom can be accounted for by several different diagnoses and, with limited to no information available from the individual, assigning an underlying cause can be extremely difficult. For example, Jiujias et al. (2017) discussed the different roles of RRBs in those with ASD and OCD, particularly in regard to anxiety. They suggested that for those with OCD, obsessions and RRBs cause increased anxiety, while for those with ASD, RRBs are associated with alleviation of anxiety. Morgan (2006) noted that behaviours such as social withdrawal, RRBs, ritualistic or compulsive behaviours, and atypical attention may be observed in individuals with ASD as well as in individuals experiencing anxiety.

Kerns et al. (2017) particularly noted not only the diagnostic overlaps in symptomology between ASD and anxiety, but also the ambiguous anxiety-like symptoms that are specific to those with ASD. Specific sources of anxiety for individuals with ASD included changes in routine, anticipation of change, anxiety around a lack of clear social rules and expectations, and intense fears of odd items/experiences or specific phobias. There is a lack of clarity around where these symptoms belong diagnostically, as they describe both ASD and anxiety.

Symptoms of Anxiety

In considering the possibility of comorbid anxiety with ASD and ID, it is also necessary to consider whether the current definition and diagnostic criteria of anxiety even make diagnosis plausible. Witwer and Lecavalier (2010) reported that comorbid psychiatric diagnosis was significantly impacted by both IQ and language skills within an ASD population, suggesting that modification of the diagnostic criteria would be necessary for accurate assessment.

In anxiety disorders, anxiety is the “future-orientated mood state” that prepares an individual for possible negative events in the future (Craske et al., 2009). Persistent, uncontrollable worry and expectation about future events defines most anxiety disorders, such as excessive apprehensive expectation for GAD (DSM-5; APA, 2013). Witwer and Lecavalier (2010) reported that when testing a population of children with ASD with and without a co-occurring ID, those with an ID reported far lower levels of “worry”, resulting in a much lower GAD diagnosis rate (8%), while those with ASD and no ID reported much higher rates of worry and 50% received a diagnosis of GAD. Given the considerable impact of co-occurring ASD and ID on an individual’s ability to predict outcomes and see the larger picture in a situation, it appears unlikely that these individuals would meet diagnostic criteria for an anxiety disorder.

It is also possible that atypical behaviours and observable behaviour patterns may be a useful way of identifying an underlying psychopathology. Tantam (2000) suggested that perseveration and increased RRBs were markers of developing a comorbid psychiatric disorder in individuals with ASD. It is therefore necessary to consider the behavioural equivalents of diagnostic criteria, rather than assuming that the same cognition-based criteria will be appropriate for all populations. Witwer and Lecavalier (2010) suggested that some individuals with ASD do not meet criteria for a valid comorbid psychiatric disorder due to the inability of a clinician to identify and assess all symptoms. They proposed that the co-occurrence of ASD with ID has a significant impact on the clinician’s ability to assess comorbid psychopathology.

Assessment Tools

Another obstacle to accurate comorbid diagnosis of anxiety is the availability of appropriate assessment instruments (Bakken et al., 2010). Given that traditional diagnostic interviews are less accessible for individuals with ASD and ID, parent or carer feedback through measurement tools are typically used as a replacement in both clinical and research settings (Helverschou et al., 2009). This practice has limitations as parents and carers may be ill equipped to accurately identify and attribute behaviours and physiological responses to their child’s internal mental state (Simonoff et al., 2008).

Bailey and Andrews (2003) likewise noted that relying solely on carer feedback can be difficult as a carer's experience of life may differ from that of the individual with ID. This difference in experience can make it problematic to attribute cause to behaviour or to identify the difference between pathological and adaptive behaviours.

A range of assessment measures have been developed to aid both diagnosis of and research into comorbid psychiatric conditions, as well as some that are anxiety specific. However, given the extensive issues with symptom overlap and differential diagnosis, it is important that the measurement tool used takes into account the unique profile of ASD and anxiety (White et al., 2009). Grondhuis and Aman completed a review of the available assessment instruments in 2012 and found that there was no current gold standard for the assessment of anxiety in individuals with ASD; a review by Moskowitz, Rosen, and colleagues in 2017 found similar results. Grondhuis and Aman (2012) have suggested that the lack of solid reliability and validity studies for many of the assessment tools raises questions about some of the prevalence rates historically reported. This could account for some of the discrepancy in reported comorbidity.

Kerns and colleagues have made adaptions to the Anxiety Disorders Interview Schedule-Child/Parent: Parent version (Silverman & Albano, 1996), namely the Autism Spectrum Addendum (ADIS/ASA; Kerns et al., 2014), in an attempt to address the symptom overlap and difficulties differentiating between ASD and anxiety. Research into the use of this measure has found it useful to consider both the traditional anxiety diagnostic categories and the ASD-specific ambiguous anxiety symptoms when considering the comorbidity of ASD and anxiety (Kerns et al., 2017).

Moskowitz, Rosen, et al. (2017) in their review of anxiety assessment tools for individuals with ASD noted the research emphasis on the "higher functioning" end of the spectrum and that the nature of many of the self-report anxiety questionnaires currently available would exclude those with a significant ID by nature of the cognitive and language load required for completion. They also pointed out the specific difficulties of dissecting anxiety from ASD behaviours, specifically RRBs, which are often grouped together on questionnaires. To date, measurement tools have not specifically taken into account the relationship between ASD and ID and the way this may affect the manifestation of anxiety. Given that Matson and Shoemaker (2009) made the point that ASD and co-occurring ID present differently than either disorder alone, this would be a necessary consideration in the development and validation of an anxiety assessment tool for this unique cohort.

MacNeil et al. (2009) completed a literature review focusing on the diagnosis of anxiety in children and adolescents with ASD. In this thorough review, they discussed the limited measures to assess anxiety available for use with the ASD population and recommended that for any measure used to assess anxiety for an individual with ASD to be valid, specific ASD normative data should be available. The authors advocated for a multimodal method in diagnosing anxiety in those with ASD, recommending that anxiety measures from multiple sources should be used in conjunction with direct observation and a clinical interview. The review also discussed the differential presentation of anxiety when considered with ASD symptom severity but did not discuss the effect of co-occurring ID and how this in turn impacts both manifestation and identification of anxiety in these individuals.

Matson and Cervantes (2014) completed a review of research focusing on the co-occurrence of ASD and psychopathology. In their review of 72 papers published between 2000 and 2013, the majority did not include individuals with ID and there was also little consistency in terms of measurement tools used. The majority of tools utilised were developed for a non-ASD population and have little to no information regarding ASD norms. While some ASD-specific measures have been developed and were reported on, there was little agreement across research groups as to which measure should be used.

Clear definitions, diagnostic criteria, and methods are therefore not readily available when considering the way anxiety manifests in individuals with ASD and ID. Mainstream definitions, including those in the DSM-5, could potentially exclude diagnosis of an anxiety disorder in many individuals with ASD and ID when they do in fact exist; this would, in turn, result in a lack of appropriate treatment. In addition, while concise diagnostic boundaries do not currently exist between anxiety and anxiety disorders, behavioural changes over time seem to offer some insight into how these phenomena may be conceptualised and defined.

Anxiety Disorders, ASD, and the DSM-5

The changes in DSM-5 diagnostic criteria have had a significant impact on how anxiety can be diagnosed for those with ASD. Under the DSM-IV (and its later text revision DSM-IV-TR), several comorbid diagnoses were not possible, particularly GAD, social phobia, or separation anxiety disorder (APA,1994, 2000). However, the reformulation of several diagnostic criteria, including for ASD and ID, has resulted in the option of diagnosing these comorbidities (DSM-5; APA, 2013).

In the DSM-5, a diagnosis of ASD now comes with a number of specifiers, including whether there are co-occurring conditions such as ID, language impairment, associated genetic or medical conditions, or associated other neurodevelopmental, mental, or behavioural disorders (APA, 2013). Professionals are therefore encouraged to consider any comorbid conditions that might also be present for the individual, which can now include all anxiety disorders. This is a considerable change in expectation, with no such specifiers being included in the DSM-IV or the DSM-IV-TR (APA, 1994, 2000).

This change encourages a more holistic approach when considering a diagnosis of ASD and possible comorbidities, which may in turn result in different treatment or program choices for the individual. Certainly, acknowledgement and understanding of issues other than ASD may alter the treatment pathway followed and the resources used to best support their learning and development.

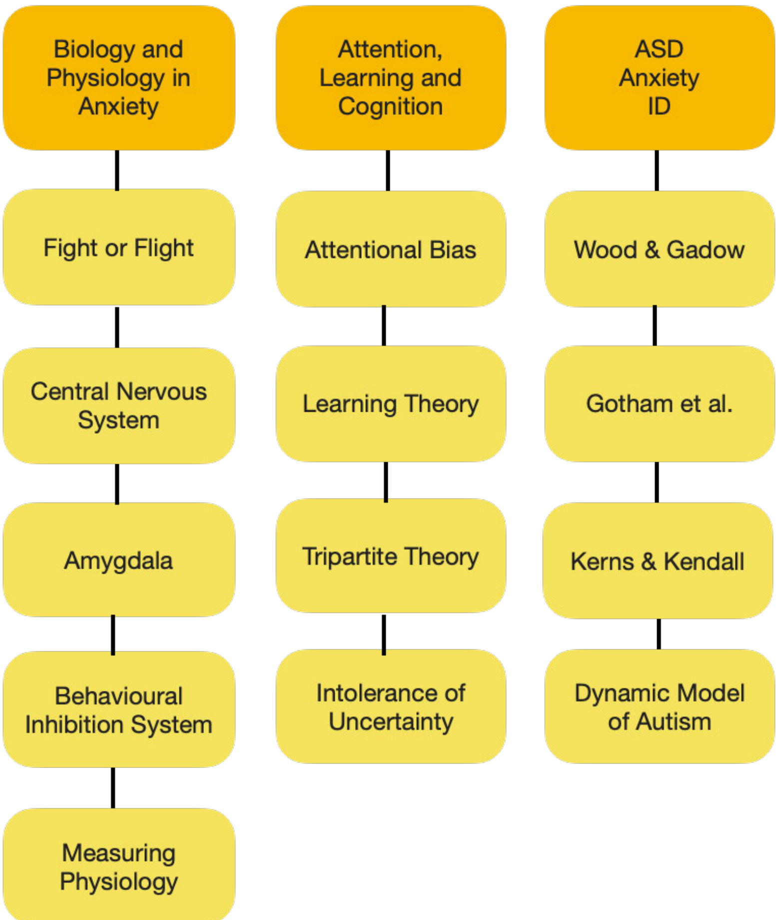
There is a myriad of challenges inherent in the identification of anxiety in individuals with co-occurring ASD and ID. Difficulties with symptom amplification, diagnostic overshadowing, and the intrinsic communication and information processing deficits associated with both ASD and ID make the identification and diagnostic process complicated and murky. The anxiety measurement tools currently available do not have norms for individuals with ASD and ID, while the tools themselves show a considerable lack in consistency in the the domains considered and items used to measure anxiety. This makes generalisation and conclusions about anxiety hard to draw across such a heterogenous population as those with ASD and ID. Despite these limitations, it is useful to consider a theoretical framework for the development of anxiety in individuals with ASD and ID, and how this may offer insight into the diagnostic process. In Chapter 4, anxiety theories created for both mainstream and ASD populations are overviewed and considered in relation to the most appropriate for inclusion in this research.

Chapter 4: Theoretical Framework for Anxiety

Anxiety and its associated vulnerabilities (both in terms of susceptibility and maintenance) have been considered from a variety of disciplines, resulting in numerous theoretical frameworks being available from which to consider anxiety. The theories discussed here have been categorised as biological/physiological, learning, attention and cognitive, and then those that are ASD specific in their design, as outlined in Figure 3. Aside from those designed specifically for the ASD population, all the theories discussed have been developed for the mainstream or neurotypical population; their applicability to those with ASD and ID will be presented. Following consideration of these frameworks, given the complex relationship between ASD, ID, and anxiety the most appropriate theoretical framework for this thesis will be discussed.

Figure 3

Overview of Theoretical Frameworks



Biology and Physiology in Anxiety

A diagnosis of any anxiety disorder under the DSM-5 requires the presence of physiological as well as cognitive symptoms (APA, 2013). Physiology is therefore a prime component of the experience of anxiety, and several hypotheses have been posited as to an individual's susceptibility, development, and maintenance of anxiety and how physiology or even underlying biology may be part of the system. These theories are explored here, including the way physiological responses can be measured, and their role as an objective option for the identification of anxiety in those with ASD and ID is discussed.

Fight or Flight

When faced with a challenge or threat, the human body has a complex sequence of hormone and neurotransmitter releases that prepare an individual to respond behaviourally and physiologically. The response is carried out by two systems, controlled by the hypothalamus: the sympathetic-adrenal-medullary (SAM) and the hypothalamic-pituitary-adrenal (HPA). These systems appear to be activated independently in response to the stressor itself. The precise roles these two systems play in responding to stressors and the way they coordinate is unclear, though they do seem to interact at several levels in cooperative, sequential and, at times, even opposite ways (Godoy et al., 2018). However, each system does seem to be linked to a different set of behavioural responses. The SAM system is primarily involved in "fight or flight" responses, or a "defence reaction". The activation of the SAM system allows for a rapid, physical, but short-lived response to a stressor. By comparison, the HPA system has a slower response; it is involved with the immune system and the effects on target tissues may not become obvious for up to an hour after the stressor. This has been described as a "defeat reaction" and is far more passive than the SAM response, characterised by behavioural withdrawal or avoidance. Research has found that while the SAM and HPA systems are activated as a result of the perception of the stressor, this assessment is highly influenced by the individual's understanding of events and the amount of control they can exert (Bauer et al., 2002).

A link has been found between hyperactivity of the HPA system and the experience of chronic anxiety disorders such as post-traumatic stress disorder (Fries et al., 2005). These researchers suggested that chronic activation of the HPA system results in high stress sensitivity, pain, and fatigue and accompanies abnormally low cortisol levels (an indicator of HPA activation). This suggests that in the face of chronic anxiety, the HPA system can struggle to recalibrate, continuing to alter the individual's responses to anxiety even after the direct stressor is no longer present.

Bitsika et al. (2014) tested this hypothesis on a group of boys with ASD but no ID. They found that self-reports of anxiety were correlated with abnormally low cortisol levels, suggesting those boys showed similar physiology to those with chronic anxiety disorders. Whether this altered cortisol level represents a predisposition within those with ASD or is indicative of their sensitivity to experiencing chronic anxiety is unclear.

Bauer et al. (2002) discussed the role of allostasis. This term is used to describe the body achieving stability following the activation of physiological systems (such as HPA and SAM) as a result of a stressor. As opposed to homeostasis and the return to a fixed point of stability, allostasis allows for ongoing fine-tuning across multiple physiological systems in response to the demands of the context and the needs of the individual. Exposure to chronic stress or arousal places considerable demand on the physiological system, which is termed "allostatic load". Chronic allostatic load can result in dysregulation of the systems, contributing to susceptibility to both physical disease and psychiatric disorders. This is particularly true if the stress systems have been excessively activated over a long period of time. In this way chronic stress, or the perception of chronic threats and challenges, may change the system's ability to respond appropriately to stress demands and lower an individual's ability to regulate their stress responses. This may explain the long-term maintenance of anxiety for individuals with ASD, as ongoing threat perception and chronic stress reduce their ability to regulate ongoing stress and anxiety responses.

Central Nervous System

Kagan et al. (1988) postulated that there are physiological correlates for behaviour problems in children, which could then be related to their anxiety and stress responses. This theory considers certain children to have an innate tendency to over-arousal of the central nervous system caused by a lower activation threshold in the limbic-hypothalamic system. This results in a specific behavioural phenotype typified by excessive fearfulness, withdrawal, and avoidance which, in extreme forms, could have long-term implications for a child's temperament and socialisation style. The antithesis would be a child who appears hyperactive, intrusive, aggressive, and lacking fear, and who would have an under-reactive nervous system with a high activation threshold by comparison. This model does not directly consider ASD, and one of its limitations is its inability to consider such comorbid behavioural conditions or states (Bauer et al., 2002). However, it does offer a possible correlate for sensory over-responsivity or emotional regulation deficits, as it considers a physiological predisposition for possible individual responses to stress.

Amygdala

The role of the amygdala in moderating fear, stress, and anxiety responses is well documented in both human and animal studies (Morgan, 2006). The amygdala is part of the limbic system and appears to be specifically involved in threat perception. It also plays a role in evaluating emotional tone during social interactions. There is some, though to date inconsistent, evidence to suggest that abnormalities of the amygdala have been seen in some individuals with ASD (Sweeten et al., 2002), and, further, that these abnormalities are similar to those produced by chronic stress resulting in increased anxiety in rodents (Morgan, 2006). The direction of this relationship is unknown; that is, does amygdala damage result from the experience of chronic anxiety or create a predisposition to be overactive to anxiety responses? While possible similarities between the neuroanatomy of an individual with ASD and an individual without ASD but with chronic anxiety are not fully understood, they suggest a specific vulnerability for individuals with ASD to experience anxiety (Morgan, 2006).

Behavioural Inhibition System

Gray (1985) has suggested a biological model to explain individual vulnerability to anxiety, based on the nervous system circuit termed the behavioural inhibition system (BIS). The BIS is anatomically located within the septo-hippocampal system, and its primary role is to evaluate environmental stimuli against expected stimuli. In this way, stimuli that are defined as aversive, for example, threats of punishment or failure, or that are novel in nature, can result in activation of the BIS, leading to current behaviour being stalled, increased arousal, and hypervigilance. An overactive BIS can therefore be considered as an underlying vulnerability to fear and anxiety development.

Overactivity of the BIS as an underlying vulnerability is a possibility for individuals with ASD, ID, and anxiety, particularly in regard to novel stimuli. As described previously, insistence of sameness and difficulty with change and new events are frequently characteristic of ASD; therefore, an internal system that is sensitised to responding negatively to novel stimuli may help explain some of this reactivity.

Measuring Physiological Changes

As physiological responses play a key role in the manifestation of anxiety, the measurement of physiological responses may offer an objective method for the confirmation for the experience of anxiety (Morgan, 2006). Friedman and Thayer (1998) reported that heart rate has been used effectively to study anxiety, such as panic disorder, as episodes involve heart palpitations. They postulated that heart rate variability (HRV) can be an indicator of the system's ability to regulate itself after experiencing stress or anxiety and to modulate resources in terms of attention and emotions. Hollocks et al. (2014) used HRV as, unlike simple heart rate, it is a measure of the system's regulation and is less affected by cognitive reasoning. Friedman and Thayer reported that low levels of HRV have been associated with emotional regulation difficulties and poor responsivity in children. Kagan et al. (1988) concurred that the children in their longitudinal study, who started as shy and inhibited 2-year-olds, later showed higher levels of unusual fears than their uninhibited peers and low levels of HRV as their system cannot regulate well. Friedman and Thayer therefore postulated on the basis of this relationship that those with a low HRV are at increased risk of developing an anxiety disorder in adulthood. As such, heart rate and the ability to regulate heart rate when faced with anxiety offers a potential indicator of anxiety and a potential early warning sign in children.

Moskowitz et al. (2013) used an electrocardiogram to collect heart rate and respiratory sinus arrhythmia (RSA; similar to HRV) data as physiological correlates for anxiety in a case study of individuals with ASD. They found that, when in situations of high personal stress, individuals with ASD showed increased heart rate, reduced RSA, and increased problem behaviours. They also found that with the introduction of stress- and anxiety-reduction strategies, these physiological responses to specific situations could be significantly reduced, along with the problem behaviours (Moskowitz, Walsh, et al., 2017). This indicates that heart rate and RSA/HSV may offer a reasonable correlate to stress and anxiety.

Morgan (2006) reported that while skin temperature and muscle tension have been used in biofeedback to aid individuals in monitoring their own stress levels, there is little evidence to support the theory that skin temperature variations occur to a level that offers a reasonable indication of internal stress. Romancyzk and Gillis (2006) reported that galvanic skin conductance is the most commonly used measure of physiological arousal. Skin conductance is directly related to sweat gland activity, which is highly correlated with arousal levels.

Cortisol is one of the hormones produced as a result of activation of the HPA system and is therefore directly involved in the experience of stress. As cortisol is part of the slower release HPA system, it typically peaks 20–30 minutes following the introduction of a stressor, at which point a reading can be taken to see the difference from baseline. Cortisol readings can be taken either through blood tests (themselves often a stressor, especially in children) or through saliva collection, requiring an individual to hold a cotton ball under their tongue for 2 minutes (Romancyzk & Gillis, 2006). More recently, diurnal cortisol collection has been utilised to test the changes in cortisol levels between morning and evening, as there is a natural pattern of decay that exists in everyone (Van Ryzin et al., 2009).

Research has focused on looking at changes in cortisol levels across the day and the way this relates to experiences of chronic and acute stress as well as behavioural manifestations. Dykens and Lambert (2013) reported that the majority of mothers with children with ASD show an overall blunted cortisol trajectory, which was also associated with lower physical health and higher levels of reported personal stress. This pattern of blunted cortisol—hypocortisolism or lower cortisol activity—is associated with exposure to chronic stress. Hypercortisolism, by comparison, is consistently high cortisol activation and often associated with acute stress. Similar results of blunted diurnal cortisol levels have been found in young children in the foster care system, particularly for those who had experienced a history of maltreatment (van Andel et al., 2014). The researchers found that this cortisol pattern could be normalised with treatment (behavioural support for the foster family) and were associated with behavioural improvement. Blunted diurnal cortisol was also found in children attending lower quality child care centres, prompting the authors to suggest that this could leave children at risk of long-term neurological and developmental outcomes (Sims et al., 2006).

It is therefore clear that physiological responses to stress may be a useful tool in objectively identifying stress or anxiety in an individual with ASD. However, the collection of such data is problematic in this population, and could in itself cause considerable stress, confounding the data being collected. Moskowitz et al. (2013) and Moskowitz, Walsh et al. (2017) found that in order for the participants to tolerate the electrodes for the electrocardiogram to be administered, extensive prior training was required. This would be a considerable limitation given the extreme variability in the sensory profile of individuals with ASD and would necessarily exclude those who could not tolerate the process, as the process itself could also provoke increased stress and anxiety. Similar collection issues exist for measures of galvanic skin conductance, blood tests, or mouth swabs for cortisol, with an individual with ASD's questionable ability to follow the tight collection protocols.

A number of theories have been posited that explain how individuals may either have a susceptibility to experiencing anxiety, or how their system may actually alter as a result of exposure to chronic anxiety. There are similarities between some individuals with ASD and individuals experiencing chronic anxiety. Given our current lack of clarity into the complexities of the central nervous system, HPA, and SAM, while these theories offer possibilities, they do not yet explain the complicated relationship between ASD, ID, and anxiety. While physiological measures offer a possible quantifiable measure of anxiety, our ability to access these measures for those with ASD and ID is extremely limited.

Attention, Learning, and Cognition

Theories of attention and learning have been developed to explain why some individuals develop anxiety and some in similar or identical situations do not. These theories focus on how information from the environment and context is observed, attended to, and processed as threat versus non-threat. These theories are explored below and considered in terms of their relevance to individuals with ASD and ID.

Attentional Bias Theories

In considering how anxiety disorders evolve and are maintained, attentional bias in processing threat information is given a prominent role (Bar-Haim et al., 2007). It is hypothesised that anxious individuals are more sensitive to, and therefore that they increasingly attend to, threat-related stimuli in the environment. Beck (1979) suggested that anxious individuals attend to threat cues more readily, interpret benign stimuli as threating, and are more likely to remember stressful experiences. Eysenck and Byrne (1992) proposed that both pre-attentive and attentional biases exist for anxious individuals, resulting in them attending not only to threat stimuli more, but by extension to the entire environment. resulting in hypervigilance. In this way, an anxious individual could constantly be scanning for possible threats and when a possibility is identified, their attention then narrows into a spotlight on stimuli.

J. Williams et al. (1997) further hypothesised that anxious individuals not only attend to threat stimuli during early preconscious stages of processing, but that in later stages they tend to direct their focus away from threat. As a result, their anxiety is increased through initial automatic response to possible threat stimuli, and subsequent avoiding of the stimuli when more cognitive processes are available to process the threat, hence denying themselves the opportunity to further evaluate the stimuli and potentially reduce their anxiety. This is similar to a panic response, which is an automatic response in nature but can become anticipated and dreaded, leading to a panic disorder when maladaptive behaviours to avoid further panic attacks occur. Maladaptive behaviours include avoidance of perceived onset stimuli such as concern that a panic attack was caused by being in a public space, so avoiding all public spaces subsequently.

The cognitive-motivational view of anxiety posits that there are two distinct motivation systems controlling cognitive and behavioural responses—valence evaluation and goal engagement systems—which together effect the individual's pre-attentive processes and initial orientation to threat stimuli (Mogg & Bradley, 1998). The valence evaluation system assesses stimuli threat, taking into account a number of factors including the nature of the threat, its context, current arousal levels, and previous experience. Anxious individuals will have a highly sensitive system, resulting in low-threat stimuli being evaluated as having high-threat values. The output of this assessment process then feeds into the goal engagement system, which is responsible for the allocation of resources to both action and cognitive processing. If a stimulus has low valence, then it can be ignored, and current goals can be pursued. However, if the valence is high then the system interrupts current activity, triggering a response to the threat, increasing orientation and cognitive processing (Mogg & Bradley, 1998). In this way, anxiety is associated with negative/high valence appraisal and external goal engagement; individuals who are vulnerable to anxiety are more likely to be future orientated and externally focused, displaying hypervigilance and anticipating possible danger. This combination of valence and goal engagement can result in a pre-attentive bias for external threat cues.

Bar-Haim and colleagues (2007) completed a meta-analytic study of the research to date regarding attentional bias in anxious individuals. Their conclusion was that there does appear to be an attentional bias in anxious individuals for threat-related stimuli when compared to non-anxious individuals. Interestingly, they also concluded that this pattern was for specific threat levels. That is, anxious individuals were biased towards mild levels of threat, while non-anxious individuals were biased towards high levels of threat. Bar-Haim et al. (2007) also found that both subliminal and conscious perception were involved in the threat-related bias.

These findings are interesting when considering how attentional bias may be affecting the development of anxiety in individuals with ASD and ID. It could certainly be the case that biases towards threat stimuli exist for individuals with ASD and ID, especially as at least part of this threat assessment appears to occur prior to cognitive appraisal, meaning that limitations in cognitive processing do not preclude the development of anxiety in this population. In fact, it could be that the cognitive difficulties experienced by individuals with ASD and ID are limiting their ability to accurately appraise their threat assessments and therefore to alter their response accordingly. In line with J. Williams and colleagues' (1997) suggestion of attention movement away from the source of threat, not only could these individuals be avoiding the opportunity to more cognitively process the threat, but they also have limited cognitive abilities to rationalise and therefore diminish their stress.

Learning Theory

Modern learning theory posits that our early learning experiences impact our response to later life stressors, which can then lead to the development of an anxiety disorder (Mineka & Zinbarg, 2006). This early learning commonly results in the development of fears or specific phobias, which Rachman (1977) suggested can be learnt in three different ways: through direct negative conditioning, through observing others' experiences, and through verbal information. This early conditioning directly impacts how an individual will interpret and respond to stressful events, particularly in regard to their perceptions and experiences with control and predictability.

Mineka and Zinbarg (2006) proposed a model that illustrates how individual vulnerabilities and learnt responses to stress can result in anxiety or panic. When faced with stressful events, an individual's perceptions of the controllability and predictability of the event, together with their learnt experiences about stress in general and the specific stressor in question, will interact and can result in either anxiety or panic. Post-conditioning responses, such as re-evaluation and presence of inhibitory conditioned stimuli, will then affect the quality and intensity of the panic or anxiety response. The authors suggested that this model can account for the development of different anxiety disorders including specific phobias, GAD, and post-traumatic stress disorder.

Of particular interest in considering how learning theory is relevant to the ASD and ID population is the distinction Mineka and Zinbarg (2006) make between panic and anxiety. They suggested that panic and anxiety are two separate factors, with panic characterised by a "strong autonomic arousal, extreme fear and fight-or-fight action tendencies and anxiety is accompanied by apprehension, worry and tension" (p. 16). This definition of anxiety carries a high cognitive load, which may prove a limitation for those with ASD and ID. It may be appropriate to consider the experience of extreme stress in those with ASD and ID as being closer to this definition of panic, rather than traditionally defined anxiety, when considered in light of learning theory.

Perception of uncontrollability and unpredictability is another interesting concept when considering those with ASD and ID. The information processing difficulties associated with ASD directly impact an individual's ability to see patterns in behaviour and to predict future events (Frith & Happe, 1994). This lack of predictability, especially to potentially stressful events, would certainly leave individuals with ASD more susceptible to panic or anxiety. Likewise, limited communication skills could reduce an individual's ability to express their needs and to understand those around them, and therefore could result in them experiencing less control of their world.

Tripartite Model of Anxiety

The tripartite model was originally developed for adults and suggests a common emotional and cognitive substrate shared between anxiety and depression, termed negative affect (NA; Clark & Watson, 1991). NA is defined as general psychological distress or negative mood underlying both anxiety (made unique by the presence of physiological hyperarousal) and depression (loss of interest or pleasure and general hopelessness). Cole et al. (1997) have extended this model to include children, taking into account developmental theory and postulating that the emotional and cognitive components of anxiety and depression evolve with age, changing in presentation as a function of development. They suggested that, in young children, anxiety and depression are essentially indistinguishable, labelling this construct as NA. However, as children develop, the constructs of anxiety and depression differentiate more clearly and start to represent separate conditions.

Cole et al. (1997) further developed the tripartite model by looking at the longitudinal relationship between depression and anxiety. Their research took into account the observation that anxiety symptoms were typically seen before symptoms of depression (Brady & Kendall, 1992). They found that children's anxiety ratings often predicted increases in depression ratings over time, though not in reverse. This, and later research by Cannon and Weems (2006), would suggest that, in children, anxiety and depression are simply two facets of the single condition, NA, and that anxiety is likely to develop chronologically before depression.

The tripartite theory has some relevance when thinking about the relationship between anxiety and ASD. ASD, as previously described, is primarily a neurodevelopmental disorder. Thus, the developmental trajectories of those with ASD are significantly altered in comparison to their non-ASD peers. The tripartite theory may explain why comorbid anxiety is more often diagnosed than depression, as depression is related to age and development, which is delayed for individuals with ASD. The underlying construct of NA may explain some of the symptom overlap between ASD and anxiety, representing more of an underlying stress response in the form of psychological distress, which may then develop into anxiety. The tripartite theory does not explore ASD or ID specifically, or the complex relationship between ASD, ID, and anxiety.

Intolerance of Uncertainty

Dugas et al. (1998) developed a model to conceptualise GAD in order to better understand it theoretically and also to drive clearer clinical treatments. Their model includes four cognitive variables for the development of anxiety: intolerance of uncertainty, poor problem orientation, cognitive avoidance, and positive beliefs about worry. A key feature of this model is intolerance of uncertainty, which is the assumption that uncertainty is worrisome and upsetting and that unpredictable events are negative and to be avoided, leading an individual to engage in cognitive, emotional, or behavioural reactions to minimise or avoid the stress of uncertainty. Intolerance of uncertainty has been found to be related to worry and has been the factor that can distinguish individuals with GAD from those with non-clinical anxiety or stress (Dugas et al., 1998). Research into this theory continues, including treatment that specifically addresses intolerance of uncertainty within a cognitive behavioural therapy framework (Dugas et al., 2022).

There has been some research into how intolerance of uncertainty may be a relevant factor for understanding the development of anxiety in individuals with ASD. Jenkinson et al. (2020), in their meta-review of the subject, found that both anxiety and intolerance of uncertainty levels were elevated in individuals with ASD, and that there appeared to be a moderate strength correlation between the constructs. Rodgers et al. (2012) have discussed the idea of *insistence on sameness* as being very similar in concept to intolerance of uncertainty, with individuals with ASD using RRBs to maintain sameness and predictability where possible to ameliorate anxiety, at least in the short term. As a result of the immediate reduction in anxiety, individuals with ASD could ascribe positive beliefs about the function of behaviours, ensuring their longer term use and lessening the likelihood of other strategies being utilised and developed, thus increasing the reliance on RRBs in an anxiety cycle. The authors go on to theorise that intolerance of uncertainty is the mediating link between atypical sensory function, rigid thought patterns, alexithymia, and anxiety development in individuals with ASD (South & Rodgers, 2017). So far, this theory has had a strong cognitive focus with many features, particularly the measurement of intolerance of uncertainty, excluding those with a co-occurring ID.

Attentional bias, learning, and cognition-based theories of anxiety development and maintenance offer possible explanations for how anxiety may present a specific susceptibility for those with ASD and ID, given their particular information processing difficulties. However, none of these theories can adequately address the complex relationship of how anxiety develops in individuals with ASD and co-occurring ID.

ASD-Specific Theories of Anxiety

The relationship between ASD and anxiety is not well understood, though attempts have been made to explore the relationship between these constructs theoretically. Prevalence is so consistent for comorbidity between ASD and anxiety that it has given rise to debate that anxiety may be better described as a trait or symptom of ASD than as a separate condition. These theories specifically developed with individuals with ASD in mind will be considered in light of their applicability to those with co-occurring ASD and ID.

Wood and Gadow

Wood and Gadow (2010) reviewed existing research in an attempt to understand the development of anxiety in youth with ASD. They particularly mention the difficulties with classifying anxiety as a "true comorbidity", as an aspect of ASD, or as something in between. They pointed out that while there can be considerable symptom overlap between anxiety and ASD, the difference can be more readily identified if the motivation for the actions is taken into account. For example, frequent and repetitive questions may be interpreted as echolalia or a verbal ritual for an individual with ASD, while the same behaviour may be motivated by fear and the need for information for an individual with an anxiety disorder (Helverschou & Martinsen, 2011). These different motivations for similar behaviours may help with differential diagnosis, though diagnosis still requires a professional to assign an internal motivation to an observable behaviour.

Wood and Gadow (2010) went on to conclude that as children with ASD and co-occurring anxiety are different from those with ASD and low anxiety, the two conditions must be distinct entities, at least part of the time. They also stated that heightened anxiety is associated with greater core ASD symptomology (Ben-Sasson et al., 2008; Kelly et al., 2008). Wood and Gadow have therefore suggested a causal model whereby the core symptoms of ASD can result in stressful experiences that heighten anxiety and mood dysregulation, which in turn increase the severity of core symptoms of ASD as a coping mechanism (see Figure 4).

Figure 4

Mood Dysregulation and Anxiety

Image removed

Note. From "Exploring the Nature and Function of Anxiety in Youth With Autism Spectrum Disorders," by J. J. Wood and K. D. Gadow, 2010, *Clinical Psychology: Science and Practice, 17*(4), p. 287 (https://doi.org/10.1111/j.1468-2850.2010.01220.x).

This model suggests that the core characteristics of ASD and social rejection could result in ASD-specific stressors for some individuals to experience heightened anxiety, in turn resulting in increased severity of ASD core symptoms. The model focuses particularly on social anxiety and how confusion and social rejection can be internalised to result in anxiety and social isolation. This assumes social awareness, motivation, and cognition to process experiences that may not be relevant or accessible for individuals with ID. It seems unlikely that being prevented from engaging in RRBs and aversive sensory experiences is enough to explain all aspects of anxiety development for those with ASD and ID.

Gotham et al.

Another attempt to explain this complex relationship has been offered by Gotham et al. (2013). They postulated that insistence of sameness (IS) behaviours could account for the considerable overlap in symptoms between the ASD and anxiety, namely, behavioural rigidity and resistance to change (see Figure 5). However, Gotham et al. found that anxiety and IS function as distinct constructs, as they were not strongly associated with each other or with ASD diagnostic scores. These researchers also found that anxiety was associated with other behavioural symptoms of ASD including irritability, attention difficulties, and aggression, as well as other psychiatric conditions.

Figure 5

Model of Anxiety—Insistence on Sameness Relationship

Image removed

Note. From "Depressive and Anxiety Symptom Trajectories From School Age Through Young Adulthood in Samples With Autism Spectrum Disorder and Developmental Delay," by K. Gotham, S. M. Brunwasser, and C. Lord, 2015, *Journal of the American Academy of Child & Adolescent Psychiatry, 54*(5), p. 35. (http://dx.doi.org/10.1016/j.jaac.2015.02.005).

This study included comparison groups between those with ASD and those with a non-autism-spectrum-based developmental delay, as well as individuals with an array of IQ scores, reporting a range of 10–141. Gotham et al. (2015) found that there was a significant correlation between anxiety and IQ, indicating that those with ASD and no ID experience more anxiety than those with ASD and an ID. Given the difficulties with appropriate anxiety measures mentioned in Chapter 3, it is appropriate to consider the measure used: Gotham et al. used the Child Behaviour Checklist, a measure designed for a mainstream population, and therefore potentially skewed to identify anxiety in those with high IQ or no ID, not those with an ID or low IQ.

Kerns and Kendall

Kerns and Kendall (2013) reported, in their review of research into the relationship between anxiety and ASD, that multiple models of anxiety in ASD may exist, whereby anxiety can be both related to ASD through its core features and can also manifest as a co-occurring disorder (the nature of which is dependent on developmental and external triggers). The authors cited limitations particularly in the operationalisation of anxiety across studies, especially in regard to ASD-specific definitions or measures. It is therefore a concern that reported prevalence of comorbid anxiety and ASD may be either overestimated or under-reported due to the misattribution of symptoms.

A complicated relationship exists between ASD and anxiety, meaning that anxiety cannot be easily defined as either a trait of ASD or a comorbid condition, instead playing a role as both. This complex association means that mainstream theories and treatment programs for anxiety, stress, or ASD are not adequate to fully understand anxiety's role in ASD, particularly when other co-occurring conditions such as ID are taken into account.

Further research by Kerns et al. (2014) has considered the difference between traditional and atypical definitions of anxiety and how these may be linked to different characteristics of individuals, including their ASD symptomology. Kerns et al. have suggested that traditional anxiety encompasses the usual anxiety diagnostic patterns expected according to the DSM-IV, including everyday worries and generalised and separation anxiety. These symptoms can be seen as quite separate from ASD symptoms and are most likely associated with higher IQ and language ability. By comparison, atypical anxiety does not so neatly conform with DSM criteria and may represent considerable overlap with ASD symptoms, being associated with higher ASD symptomology and possibly with higher anxiety levels. As such, atypical anxiety may manifest in a more individually driven manner and include such things as social avoidance, anxiety around changes in routine, specific phobias, and compulsive actions.

Research into this model of typical and atypical anxiety was conducted by Kerns et al. (2014) and, while it included some individuals with mild ID, those with a full-scale IQ below 60 were excluded from the study, minimising the applicability of results to those with more significant ID. The measurement tool for anxiety used was the Anxiety Disorders Interview Schedule (ADIS), a measure created for mainstream individuals and based on DSM-IV criteria for anxiety disorders. To measure atypical anxiety, an ASD-specific addendum was created by the authors to accompany the ADIS.

Figure 6

Predictors of Traditional and Atypical Anxiety

Image removed

Note. From "Traditional and Atypical Presentations of Anxiety in Youth With Autism Spectrum Disorder," by C. M. Kerns, P. C. Kendall, L. Berry, M. C. Souders, M. E. Franklin, R. T. Shultz, J. Miller, and J. Herrington, 2014, *Journal of Autism and Developmental Disorders, 44*, p. 2858. (https://doi.org/10.1007/s10803-014-2141-7).

The predictors of typical and atypical anxiety are included in Figure 6. Interestingly, there is no relationship between IQ and either typical or atypical anxiety, though as mentioned those with significantly low IQ were excluded. However, not surprisingly, language ability was related to traditional anxiety and not to atypical anxiety. Also of particular note is that ASD symptoms were predictive of atypical anxiety, but not of traditional anxiety.

This model of anxiety, including both traditional and atypical manifestations of anxiety, offers an interesting inroad to considering how anxiety may be understood for those with ASD and ID. There is significant overlap between several of the factors offered by Kerns et al. (2014); for example, hypersensitivity is listed under the DSM-5 as a diagnostic symptom of ASD (APA, 2013), so it is interesting that those two predictors appear to relate to separate factors. Similarly, an “anxious cognitive style” appears to have overlap with both “language ability” and RRBs (within ASD symptoms), which may explain why it correlates to both factors as well. This concept of traditional and atypical anxiety markers has considerable merit, though the factors used to identify these traits may have been limited in scope, as was the population used in the research.

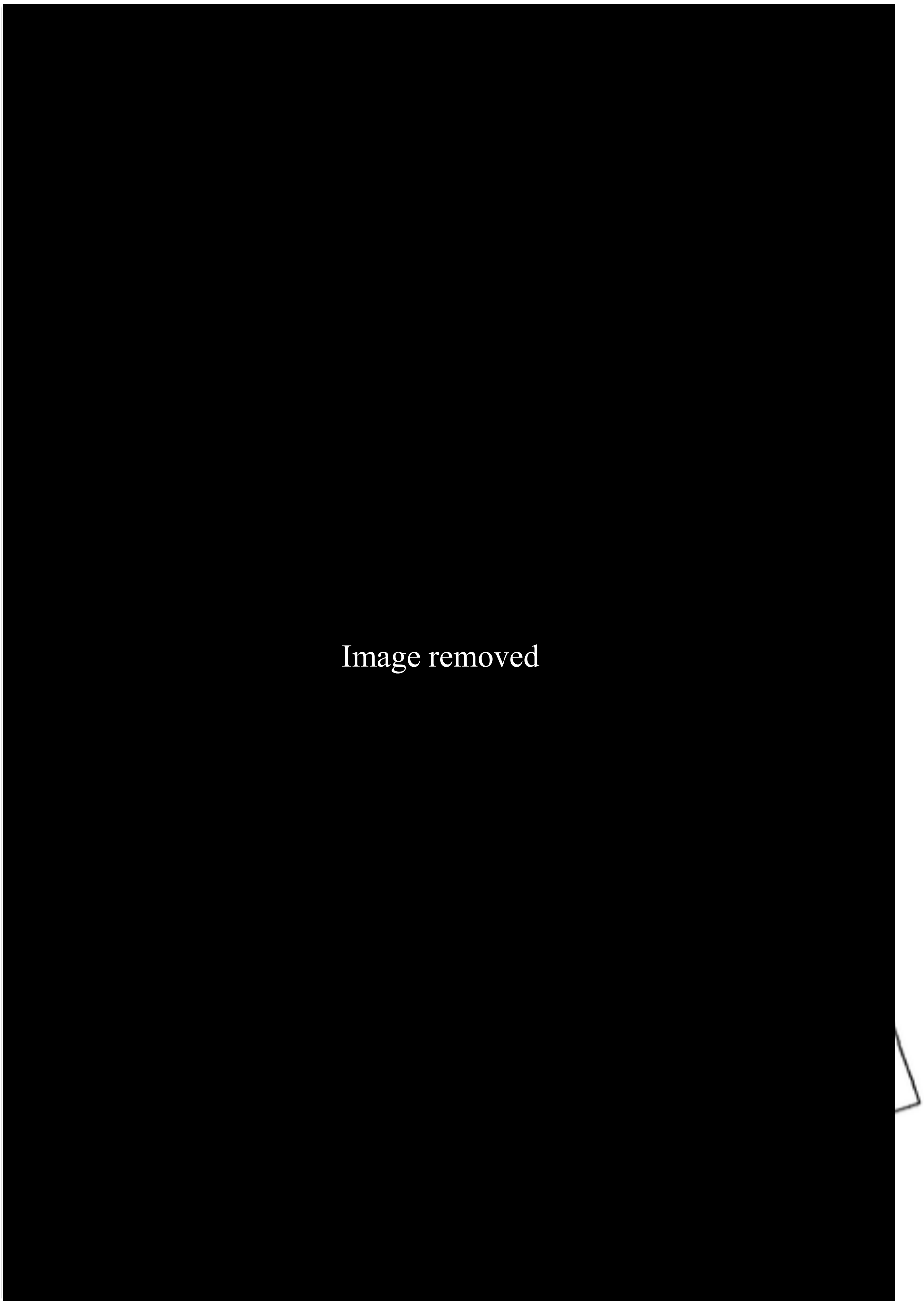
Individuals with ASD and anxiety may present in both typical and atypical ways; however, if atypical anxiety is more relevant for those with lower IQs and reduced language ability (ID), then this represents a significant reason as to why traditional diagnostic techniques and criteria are not identifying anxiety in those with ASD and ID.

Dynamic Model of Autism

Another model of anxiety development in individuals with ASD has been offered, one that attempts to account for the complex relationship between ASD and anxiety without reliance on cognitive abilities: the dynamic model of autism (Bartak et al., 2006; see Figure 7). This model considers the two clusters of ASD characteristics separately as well as how they change over time. That is, repetitive or ritualistic behaviours show varying patterns of development, while deficits in communication and socialisation are more likely to persist chronically into adulthood.

Figure 7

Dynamic Model of Autism



Note. From "Therapist Insights in Working with Stress in People with Autism Spectrum Disorder," by L. Bartak, V. Bottroff, and J. Zeitz, 2006, in M. G. Baron, J. Groden, G. Groden, and L. Lipsitt (Eds.), *Stress and Coping in Autism*, p. 249. Copyright Oxford University Press.

Bartak et al. (2006) postulated that the primary deficits of ASD are the individual's ability to process information, specifically in three areas: (a) about other people's emotions, motives, and behaviour in social interactions; (b) receptive and expressive communication; and (c) executive functioning, due to difficulties with central coherence. When an individual with these deficits then interacts with others or experiences environmental stressors, the secondary features of ASD develop as a means of coping with the associated stress. That is, repetitive and ritualistic behaviours and even sensory integration issues may develop as a means of trying to control or understand the unpredictable environment around them. Simultaneously, as social communication abilities are impacted, odd social behaviours may cause the individual to become increasingly withdrawn and avoidant of social contact.

The model goes on to suggest that when stress continues to be experienced by an individual with ASD, including poorly considered intervention that focuses only on the secondary features of ASD, the outcome will be an increase in challenging behaviours. These tertiary level behaviours may be internal, including anxiety, poor self-esteem, and paranoia, or they may be more external, presenting as physically acting out behaviours, self-injury, or property destruction.

The quaternary level is reached when interventions and responses to the challenging behaviours at the tertiary level do not take into account the functional nature of the behaviour or how they have developed. At this stage the stress experienced by the individual may now be manifesting as psychiatric disorders including anxiety, depression, or personality disorders. As a result of this accumulation of stress, behavioural challenges, and continued communication impairments, differential diagnosis is extremely difficult and can easily result in misdiagnosis.

This model considers the unique features of ASD that appear to make an individual vulnerable to stressful situations. It also points out that should the core features of ASD not be adequately supported, this vulnerability seems to increase. The model does not rely on cognitive ability to explain the presence of anxiety, as the core features of ASD at the primary level, while information processing based, do not exclude individuals with a co-occurring ID.

When an individual has difficulty predicting the motives and behaviours of others, or perceiving patterns within the environment, and then cannot seek clarification from others, stress can result from confusion and fear of the unknown. As such, almost everything in the environment can be seen as a possible stressor for an individual with ASD. New people entering their space can be interpreted as unpredictable and therefore unsafe. New spaces and locations offer a similar unpredictability, leading to unclear boundaries and uncertain expectations for what will take place within that space. Changes to routine and schedules can also cause stress, when what was predictable and known becomes unreliable and inconsistent. This conceptualisation of the interaction between ASD and anxiety can be seen to include the typical and atypical symptoms of anxiety found by Kerns et al. (2014). This difficulty in processing information and making sense of the world can be seen as a core component of ASD; however, when the individual has the additional comprehension and problem-solving difficulties associated with a co-occurring ID, these difficulties are likely to be further exacerbated, particularly when communication skills are significantly impaired.

In considering this model, anxiety and ASD are inherently intertwined. Anxiety may be considered as a continuum related to the changing levels of stress experienced by the individual, rather than as a binary present/absent concept. In this way an individual may move along the continuum as different demands and stresses are placed on their communication and socialisation abilities, resulting in different behavioural outcomes.

Stress may be what the individual with ASD experiences when in an uncomfortable or challenging situation, while anxiety may be considered to be the behavioural changes that take place as a result of the ongoing stress. An anxiety disorder could then be seen as a chronic pattern of behavioural changes that limit an individual's quality of life. However, the exact boundaries between each of these phenomena is unclear. If stress and anxiety, and movement between the two, were considered on a continuum, then behavioural changes over time would become the best indicators of increasing anxiety. Likewise, a persistent long-term change may then meet criteria for an anxiety disorder.

Aims of the Current Research

A review of the current research into individuals with ASD and ID reveals a lack of understanding about how anxiety is identified and diagnosed in this population. The complex interplay of symptom amplification and diagnostic overshadowing for individuals with ASD, ID, and possibly anxiety, makes differential diagnosis problematic. Even ASD-specific measurement tools do not necessarily take into account the communication and insight difficulties experienced by individuals with ASD and ID. Physiological measures are not readily accessible for this population, as the data collection methods used are likely to result in increased anxiety, impacting both the validity and reliability of the process and the outcomes.

It therefore seems likely that in those with ASD and co-occurring ID, anxiety is currently being underdiagnosed and that these individuals are not being identified. Given the changes in DSM-5 diagnostic and reporting criteria, it is timely to consider the issue of identification of anxiety in the population of individuals with ASD and ID.

Various models have been proposed to explain the relationship between anxiety and ASD. The majority of the models are more applicable to individuals with normal to high cognitive ability and ASD, with only one model found—the dynamic model of autism (Bartak et al., 2006)—that applies to all individuals with ASD irrespective of ID status. A key hypothesis of the dynamic model of autism is that the core characteristics of ASD in combination with aspects of the environment result in stress for the person with autism. Interventions that do not take into account the dynamic nature and function of the relationship between ASD symptoms and stress are likely to result in increased anxiety, and then an anxiety disorder that is potentially more complex to treat.

To determine how anxiety may be affecting individuals with ASD and ID, two key concepts will be considered in this research: firstly, a theoretical framework for understanding how anxiety relates to ASD and ID, and then the possibility of using behavioural markers to identify anxiety in individuals with ASD and ID. The dynamic model of autism (Bartak et al., 2006) offers a theoretical basis for understanding the development of anxiety in individuals with concurrent ASD and ID; however, this theory does not appear to have been tested empirically with either an ASD or an ASD and ID population. It therefore offers a previously untested framework and possible new theoretical basis when considering how anxiety develops in individuals with ASD and ID. This will be addressed in Study 1(Chapter 5): Does the dynamic model of autism explain the relationship between anxiety and ASD for individuals with co-occurring ID?

Difficulties in diagnosing anxiety in individuals with ASD and ID are numerous. Issues such as diagnostic overshadowing, differential diagnosis, symptom amplification, and difficulties with measurement tools may all be leading to underdiagnosis and therefore to a lack of treatment for significant anxiety issues for individuals with ASD and ID. Consideration should therefore be given to whether behavioural patterns of anxiety exist for individuals with ASD and ID, and whether these offer a more consistent method of identification and possible diagnosis. This will be addressed in Study 2 (Chapter 6): Are there specific behavioural patterns for the expression of anxiety in individuals with ASD and ID?

Chapter 5: Study 1: Investigating the Dynamic Model of Autism

In the previous chapter, theoretical frameworks for the development of anxiety in individuals with ASD were considered, including biological, physiological, learning, and attention-based, and ASD-specific theories. However, only the dynamic model of autism (Bartak et al., 2006) allowed for the co-occurrence of ID with ASD. This theory therefore offers the most suitable framework for considering how anxiety develops in those with ASD and co-occurring ID, and was tested in Study 1.

Study 1 investigated how anxiety develops in individuals with ASD and ID utilising the theoretical framework of the dynamic model of autism (Bartak et al., 2006). This model considers why individuals with ASD are particularly vulnerable to developing anxiety; unlike other models, it does not exclude those with an ID. The model has been offered on theoretical grounds but does not appear to have been empirically tested. As it is the most inclusive theoretical framework found for individuals with concurrent ASD and ID in relation to the development of anxiety, it was chosen for this investigation of the relationship between ASD, ID, and anxiety. In Study 1 the capacity of the dynamic model of autism to account for the relationship between autism, ID, and anxiety was examined, with findings reported in this chapter.

Study 1 utilised hierarchical multiple regression analysis to address the question: Does the dynamic model of autism (Bartak et al., 2006) explain the relationship between ASD and anxiety in individuals with ASD and ID? This method was considered appropriate for the analysis, as it is based on a theoretical relationship between a dependent variable and multiple independent variables, which can be introduced to an analysis in a predetermined hierarchy (Tabachnick & Fidell, 2019). That is, Bartak et al. hypothesised that there is a progression in the way anxiety develops in those with ASD and ID, and in the way in which various independent variables become relevant to the equation. The theoretical model was tested to see how well the independent variables could account for variance of the dependent variable, in this case, anxiety (Tabachnick & Fidell, 2019). As such, the dynamic model of autism with its well laid out sequence of development was considered well suited to a hierarchical multiple regression.

The definition and diagnosis of anxiety and anxiety disorders is unclear in individuals with ASD and ID (Appleton et al., 2019), and thus careful consideration must be given to how data are collected. Anxiety can be considered as a continuum rather than as a binary concept; therefore, measurement tools need to be considered that will allow for measurement of degree of intensity to provide greater sensitivity. As previously mentioned in the scoping review in Chapter 3, no measurement tools exist that consider the unique presentation of anxiety for an individual with ASD and ID. However, two tools have been found with anxiety subscales that have been used in research with individuals with either ASD or ID: the Developmental Behaviour Checklist (DBC) and the Nisonger Child Behaviour Rating Form (NCBRF). These subscales focus on different markers for anxiety: the DBC utilises behavioural markers, while the NCBRF has more performance- and language-based items on its anxiety subscale. There is little crossover or similarity between these two anxiety subscales, as is evident when considering the items for each subscale (see Table 6).

Table 6

Individual Items on DBC and NCBRF Anxiety Subscales

DBC Anxiety	NCBRF Anxious
Cries easily for no reason, or over small upsets	Overly sensitive – feelings easily hurt
Distressed about being alone	Exaggerates abilities or achievement
Excessively distressed if separated from familiar person	Feelings easily hurt
Fears particular things or situations e.g., the dark, insects	Feels others are against him/her
Fussy eater or has food fads	Feels worthless or inferior
Loss of appetite	Nervous or tense
Shy	Overly anxious to please others
Upset and distressed over small changes in routine or environment	Says no one likes him/her
	Self-conscious or easily embarrassed
	Shifts rapidly from topic to topic when talking
	Steals*
	Sulks, is silent and moody
	Talks too much or too loudly
	Too fearful or anxious
	Worrying
	Argues with other children or peers ^

Note. *Question appears in NCBRF Parent only; ^question appears in NCBRF Teacher only.

Both the DBC and NCBRF can be completed by parents and teachers. Lecavalier (2006), in his study of behaviour problems in children with pervasive developmental disorders, used the NCBRF with parents and teachers and found a significant difference in the data collected from parents compared to teachers, as have other researchers (Kamphaus et al., 1997, 1999; Llanes et al., 2018). To capture these different views and to better encompass the range in anxiety data available, both parent and teacher data are collected in this research.

Method

Setting

All participants were students or clients at Giant Steps Australia, an ASD-specific preschool, school, and adult day program based in Sydney, with a recently opened school in Melbourne. The preschool and schools in Sydney and Melbourne have a transdisciplinary collaborative approach to teaching and intervention and follow a national curriculum-based classroom program with substantial support from occupational, speech, and music therapies and psychology. Many have attended Giant Steps Sydney exclusively throughout their schooling from preschool or kindergarten, while others have joined the school at varying ages. Giant Steps has a strong transition policy, whereby students and families are encouraged to consider other schooling options when students are ready to transition into more inclusive environments such as ASD-specific or multicategorical support classes in mainstream schools, and mainstream classrooms. As a result, there is more variability in diagnosis at younger ages at Giant Steps, with most older students diagnosed with severe ASD (i.e., requiring substantial support in daily life) and moderate to severe ID. The majority of students are functionally non-verbal, and all students use a variety of augmented and alternative communication devices and supports, including visual supports (such as daily schedules, choice board, and visual calendars), picture exchange communication systems (PECS), iPads, and scripting.

The adult program is a community participation program for school leavers. Programs are individually created for the adults and can include a combination of work experience, community access, arts programs (including drama, music, and art), and activities to develop daily living skills. All the participants in the adult program have a diagnosis of severe ASD and moderate to severe ID and require very substantial support across multiple contexts.

An ASD-focused organisation such as Giant Steps facilitates consistency of schooling program and therapy support. Any additional therapies and interventions accessed outside of the school or adult program during the assessment period were captured via demographic information. Anxiety-related behaviours and ASD symptomology are expected to change with development; recruitment of children and adults from across age groups provided a cross-section at various stages to allow for examination of change related to age and development.

Participants

Children and young adults with an existing diagnosis of ASD and ID were recruited through Giant Steps Australia. In 2016, 103 families were initially invited to take part in the research; 62 (60.19%) completed the questionnaires themselves and 99 (96.12%) agreed for school staff to complete further data on their children. In 2018 an additional 36 families were invited to take part; 10 families (27.78%) completed the questionnaires themselves and 20 (55.56%) agreed for school staff to collect data about their children. In total, this provided data for 119 participants: 72 (52.18%) parent responses to the questionnaires (4 of which were parents only, 68 were shared with staff responses) and 115 participants (83.11%) with staff responses (54 staff members completing these responses) for the same set of individuals with ASD and ID. Of these, 105 participants attended the Sydney school and 14 attended the Melbourne school.

Participants met the following criteria:

1. Existing clinical diagnoses of ASD and ID (with psychometric and paediatric/psychiatric reports to support this).

2. Currently enrolled at Giant Steps preschool, school, or adult program at either the Sydney or Melbourne campus.

Participants were not excluded based on level of ID or level of expressive or receptive language.

Participant information is set out in Table 7. Of the 119 participants, 66.39% were male, 70.42% were school aged (6-18 years old), 98.32% had an ID diagnosis and 89.92% had a moderate to severe ID. In terms of comorbid diagnoses, 44.54% of the population had a diagnosis of anxiety with 26.05% having another diagnosis (e.g., ADHD). Daily medication for treating anxiety was reported in 39.50% of the population.

Table 7

Participant Information

	Number $N = 119$	Percentage %
Gender		
Male	79	66.39
Female	40	33.61
Age		
4–5 years	22	18.50
6–11 years	43	36.13
12–18 years	42	35.29
19–27 years	12	10.08
Level of ID		
No ID	2	1.68
Mild	10	8.40
Moderate	60	50.42
Severe	47	39.50
Comorbid diagnosis		
Anxiety	53	44.54
ADHD	16	13.45
Depression	11	9.24
Bipolar	4	3.46
Medication		
Daily anxiety medication	47	39.50
Daily medication for conditions other than anxiety (e.g., epilepsy, insomnia)	30	25.21

Information about the respondents completing the questionnaires, that is, parents and staff from the school, is outlined in Table 8. The parent questionnaires were completed by 88.89% mothers; parents were 93.06% English speakers and 58.33% had graduate or postgraduate degrees. The staff questionnaires were completed by 72.22%

females; 83.33% were teachers or therapists with 85.19% having a graduate or postgraduate degree and 50% having worked at Giant Steps for 5 years or more.

Table 8

Respondent Information

Informant		Number	Percentage %
Parents		(*N* = 72)	
	Relationship to child		
	Mother	64	88.89
	Father	7	9.72
	Grandparent	1	1.39
	Primary language spoken at home		
	English	67	93.06
	Other	5	6.94
	Highest educational level		
	No answer	6	8.33
	High school	15	20.83
	TAFE / Cert / Diploma	9	12.50
	Graduate or bachelor's degree	35	48.61
	Graduate degree	7	9.72
Staff at Giant Steps		(*N* = 54)	
	Gender		
	Female	39	72.22
	Male	15	27.78
	Job description		
	Teacher	18	33.33
	Occupational Therapist	11	20.37
	Speech Therapist	10	18.52
	Music Therapist	6	11.11
	Day Program Staff	9	16.67
	Length of time at Giant Steps		
	< 1 year	11	20.37
	1-2 years	5	9.26
	3-5 years	11	20.37
	5-10 years	22	40.74
	+10 years	5	9.26
	Highest educational level		
	Graduate or bachelor's degree	43	79.63
	Postgraduate degree	3	5.56
	High school	8	14.81

Materials

Measurement tools were chosen based on their emphasis on behavioural markers and history of use with ASD and/or ID populations. Data were collected to represent each variable of the dynamic model of autism (Bartak et al., 2006) as outlined in Table 9.

Table 9

Measurement Tools Used for Each Stage of the Dynamic Model of Autism

Stage and behaviour manifestation	Measurement tool
1. Primary	
Core ASD characteristics:	
Difficulties with:	*The Social Communication Questionnaire (SCQ)*
- Understanding people's feelings, motives, and behaviour	
- Receptive and expressive communication	
- Executive functioning	
2. Secondary	
Consequential ASD characteristics:	
- RRBs	1. *The Repetitive Behaviour Questionnaire (RBQ)*
- Sensory integration issues	2. *Short Sensory Profile 2 (SSP2)* and *Adolescent/Adult Sensory Profile (AASP)*
3. Tertiary	
Behavioural effects	
Challenging behaviours:	*1. The Developmental Behaviour Checklist (DBC) – Total Problem Behaviour Score (TPBS)*
Internal	*2. Nisonger Child Behaviour Rating Form (NCBRF) – Self-Injurious Behaviour subscale (SIB)*
- Anxiety	
- Poor self-esteem	
- Paranoia	
External	
- Physical behaviours	
- Self-injury	
- Destructiveness	
4. Quaternary	
Psychiatric comorbidity	
Psychiatric disorder/syndrome:	1. *DBC Anxiety subscale*
- Anxiety disorder	2. *NCBRF Anxious subscale*
- Depression	
- Personality disorder	

The Social Communication Questionnaire (Lifetime version; SCQ; Rutter et al., 2003) is a 40-item questionnaire with yes/no responses which assess social interactions and communication. Three domains are produced after scoring: reciprocal social interaction, communication, and restricted, repetitive, and stereotyped patterns of behaviours. The SCQ has both *Current* and *Lifetime* versions available. As long patterns are being considered within this study, the Lifetime version was selected as being most relevant. The SCQ was derived from the longer Autism Diagnostic Interview Revised (Lord et al., 1997) and has an autism cut-off score of 15 indicating the likelihood of an individual having ASD. The SCQ is reported to have good psychometric qualities as a screening measure, with Berument et al. (1999) reporting a sensitivity of 0.96 and a specificity of 0.80 when using the SCQ as a diagnostic discriminator. The SCQ has been used in research with children and adults and has been reported to have good internal consistency (Cronbach's $\alpha = .87$; Brooks & Benson, 2013). The Cronbach alpha was analysed for the current data set, and with the parent data was found to be .81, and with the teacher data .80.

The Repetitive Behaviour Questionnaire 2 (RBQ 2; Honey et al., 2012) is a revised 20-item questionnaire developed from the Repetitive Behaviours Interview (South et al., 2005). Behaviours are rated based on frequency or severity on a 3- or 4-point Likert scale and produce four subscales: repetitive movements, repetitive use of language, sameness behaviours, and circumscribed interests, with higher scores indicating more difficulties with repetitive behaviours. The RBQ has been used in research involving individuals with ASD and is reported to have good internal consistency (Cronbach's $\alpha = .85$; Leekam et al., 2007). The Cronbach alpha for the current data set was tested, and with the parent data was found to be .82, and with the teacher data was .72.

The Sensory Profile 2–Short form (SSP-2; W. Dunn, 2014) and the Adolescent/ Adult Sensory Profile (AASP; Brown & Dunn, 2002) were designed to assess the way individuals respond to sensory stimuli. The SSP-2, a 34-item questionnaire, is a standardised sensory measure used in research (Z. Williams et al., 2018). When tested, the Cronbach alphas for the current data set were .91 for the parent data and .82 for the teacher data, suggesting good internal consistency. The SSP-2, however, is only valid for use with individuals up to 14 years of age, necessitating the use of the AASP for older participants. The AASP has reportedly good psychometric qualities with comparison scores available for both disability and non-disability populations (Eeles et al., 2013). Cronbach alphas for the current data were found to be .89 for the parent data and .81 for the teacher data, suggesting good internal consistency.

The Developmental Behaviour Checklist (DBC; Einfeld & Tonge, 1995) was developed as a means of quantifying and monitoring behavioural and emotional problems in individuals with developmental and/or intellectual disabilities. It has two forms, the Parent (DBC-P) and Teacher (DBC-T) questionnaires, and grades the frequency of observable behaviour traits on a 3-point Likert scale, 0 = *not true* – 2 = *very true / often*, across either 96 items for the parent form or 94 for the teacher form. Results are then tabulated into an overall score of Total Problem Behaviour Score and five subscales: Disruptive/Antisocial, Self-Absorbed, Communication Disturbance, Anxiety, and Social Relating. Higher scores indicate more difficulty in those areas; results can also be compared to a normative population of individuals with the same level of ID or of the same gender. Of primary interest in this study will be the Total Problem Behaviour Score, which is the sum of all the subscales (96 or 94 items depending on parent or teacher form), and Anxiety, which is made up of eight items. The test-retest reliability is high, DBC-P = .83 and DBC-T = .76. Likewise, the internal consistency indicates good reliability (Cronbach's αn = .94; Einfeld & Tonge, 2002).

For the current data set, the Cronbach alpha was found to be .93 for both the parent and teacher data, suggesting very good internal consistency.

The Nisonger Child Behaviour Rating Form (NCBRF; Aman et al., 1996), like the DBC, was developed to assess behaviours in children and adolescents with ID and/or ASD. It has two forms—teacher and parent—that have the same set of 10 social competence items and 66 problem behaviour questions, but different scoring methods are used for each form. The questions are answered using a 4-point Likert scale regarding the frequency of such behaviours – 0 = *not true/did not occur* to 3 = *always true/occurs a lot*. Eight scores are then produced: Compliant/Calm, Adaptive/Social, Conduct Problem, Anxious, Hyperactive, Self-Injury/Stereotypic, Self-Isolated/ Ritualistic, and Overly Sensitive/Irritable. Of particular interest in this study are the Anxious subscale, with 16 items, and the Self-Injury subscale, with nine items. The NCBRF has been used in several studies and is reported to have good internal consistency, with all subscales reported to have a Cronbach alpha above .70 (Lecavalier et al., 2004), though it should be noted that it has been normed for a developmental disability population rather than an ASD-specific one (Tasse et al., 1996). The current data set was tested, and a Cronbach alpha of .90 for the parent data and .92 for the teacher data was found, suggesting very good internal consistency.

Procedure

All families with a child attending Giant Steps preschool, school, or adult program at either Sydney or Melbourne campuses were invited to take part in the study. Parents and caregivers were given information about the research project through parent information nights as well as in information sent home directly to families and through the weekly school newsletter. Parents were invited to participate in 2016 and then those who had not been enrolled at that time were invited in 2018, with the same information opportunities of parent information nights, the weekly school newsletter, and information sent directly home. Parents who declined to participate themselves were offered the opportunity for their child to be part of the data collection completed by staff at the school.

Parents were asked to complete demographic information including age, existing comorbid psychopathology, medical conditions, medication taken, schooling history, living environment (several students and young adults are in full- or part-time out-of-home care), family history of anxiety or other psychopathology, confirmation of ASD and ID diagnosis, and level of impairment.

For each student in the school, the questionnaires were divided amongst the professional staff working in each classroom, namely the class teacher, occupational therapist, speech therapist, and music therapist assigned to each class. Data collection took place in the second half of the year to ensure staff had a minimum of 6 months working directly with individuals and knew them well enough to answer the questions. Teachers completed the DBC and NCBRF to ensure consistency between the two anxiety rating forms. Therapists completed the SSP2/AASP, SCQ, and RBQ.

For the adults in the day program, data were completed by program staff who had worked with the individuals for a minimum of 6 months. There were no teachers and limited therapists within the day program, so support staff who worked with the clients daily were asked to complete the questionnaires.

Ethics

Ethical approval was sought and granted from both Giant Steps and the Griffith University Ethics Committee for this research to be conducted. The Griffith University Human Research Ethics reference number for this project is: 2016/010. These approvals are included in Appendices A and B.

Data Analysis

Descriptive information for each of the variables was completed, including investigation into normal distribution through skewness and kurtosis. The values for asymmetry and kurtosis between -2 and +2 are considered acceptable to prove normal univariate distribution (George & Mallery, 2019). As part of this investigation, two variables were found to have non-normal distribution: NCBRF (Anxious) for both the parent and teacher data, and the RBQ for teacher data. As such, the data for these variables were transformed using Statistical Package for Social Sciences (SPSS) Rank Case to fit normal distribution more closely and to be usable within the parametric statistical analysis planned, that is, the hierarchical multiple regression.

There were minimal missing data points, with six parent questionnaires missing from three students from the complete pool of 426 parent questionnaires. For these three cases, where missing data did exist, cases were excluded pairwise. There were no missing data from the staff questionnaires.

Correlation Between NCBRF Anxious and DBC Anxiety. Two subscales for anxiety were utilised in this study, each capturing different aspects of anxiety; DBC Anxiety is behaviour based and NCBRF Anxious is performance and language based. A Pearson correlation was computed between these two scales for both the parent and teacher data, to test the relationship between the subscales.

Hierarchical Multiple Regression. Harrell (2001) suggested that to have adequate predictive power, there should be at least 10 subjects per predictor. For this ratio to be met within this study, a sample size of at least 100 subjects would be needed. This is a preliminary exploratory study, with small subject groups of 72 in the parent group and 115 in the staff group. This limits the predictive power possible from the sample, particularly from the parent data, but does suggest that any significant results found would also be present within a larger population. To account for the limitations with power, significance was set at $p < .01$ to ensure robust relationships within the data would be identified and reported on.

The independent variables were entered as blocks in three steps according to the dynamic model of autism (Bartak et al., 2006) as follows:

Step 1: Age, ID level, and SCQ

Step 2: RBQ, SSP2/AASP (4 subscales)

Step 3: DBC TPBS, NCBRF SIB

Four regression analyses were conducted each following this step progression, but using a different dependent variable, namely NCBRF Anxious Parent and Teacher and DBC Anxiety Parent and Teacher.

The DBC contains a different number of items for the child and adult questionnaires, which altered the number of questions used to total the subscale scores, particularly the Total Problem Behaviour Score and Anxiety, impacting the relatability of the data between the child and adult populations. Following the advice of Taffe et al. (2008) about different ways to extract information and data from the DBC, the DBC raw scores were converted into mean item scores. This gives a measure of the intensity of each subscale irrespective of the number of items. SPSS 27 was used to run the analyses.

Results

Descriptive statistics were completed for each of the independent and dependent variables used in the analysis; an overview is available in Table 10. When these scores were checked for normal distribution, three variables were found to have kurtosis statistics outside of the acceptable range of -2 to 2. In looking at the data spread of NCBRF for both the Parent and Teacher variables, the scatterplots skew positively, while RBQ Teacher shows a positive kurtosis. These variables were NCBRF (Parent), NCBRF (Teacher), and RBQ (Teacher); these are highlighted in red in Table 10. To maintain these variables in the analysis and to continue with the planned regression, the three variables were transformed via SPSS Rank Case (see Table 10). This process of data transformation is recommended by Tabachnick and Fidell (2019), though with caveats regarding interpretation of the outcomes of the data. The updated variables, following transformation by SPSS, were used in regression calculations. Data were assessed for multivariate outliers using a Mahalanobis Distance Test (Tabachnick & Fidell, 2019). No multivariate outliers were identified.

Table 10

Descriptive Statistics of Variables

Variable	Range Min	Range Max	Mean	SD	Skewness statistic	Skewness SE	Kurtosis statistic	Kurtosis SE
Gender	1	2	1.34	.47	.70	.22	-1.53	.44
Age	4	27	11.35	5.56	.78	.22	.05	.44
ID level	1	4	1.72	.69	.74	.22	.65	.44
Parent informant								
DBC Anxiety	.00	1.78	.60	.39	.85	.28	.59	.56
DBC TPBS	.04	1.38	.62	2.45	.30	.28	.53	.56
NCBRF Anxious	.00	26	5.44	5.54	1.48	.28	2.24	.56
NCBRF SIB	.00	19	4.58	4.71	1.27	.28	1.01	.56
RBQ	10	61	35.71	9.82	-.11	.28	.48	.56
SCQ	3	36	21.74	6.50	-.50	.28	.14	.55
SSP2/AASP Bystander	7	58	23.19	10.36	.93	.28	.78	.55
SSP2/AASP Seeker	8	43	24.56	8.87	.21	.28	-.49	.55
SSP2/AASP Avoider	5	60	31.25	9.99	.17	.28	.95	.55
SSP2/AASP Sensor	11	54	33.22	8.57	-.28	.28	.36	.55
Teacher/Therapist								
DBC Anxiety	.13	1.88	.69	.36	.65	.23	.31	.45
DBC TPBS	.23	1.44	.66	.25	.88	.23	.81	.45
NCBRF Anxious	.00	39	6.25	6.71	1.94	.23	5.29	.45
NCBRF SIB	.00	23	6.32	5.27	.98	.23	.57	.45
RBQ	2	58	37.38	7.45	-.39	.23	4.18	.45
SCQ	2	34	23.72	6.05	-1.20	.22	1.76	.45
SSP2/AASP Bystander	6	59	25.99	12.62	1.06	.23	.22	.45
SSP2/AASP Seeker	12	55	25.82	8.77	.98	.23	.83	.45
SSP2/AASP Avoider	18	58	32.97	7.91	.54	.23	.28	.45
SSP2/AASP Sensor	17	48	35.35	5.92	-.48	.23	.28	.45

Transformed variables								
NCBRF Anx Parent	5	72	36.5	20.82	.02	.28	-1.23	.56
NCBRF Anx Teacher	8.5	114	57.5	32.91	.02	.23	-1.22	.45
RBQ Teacher	3	114.5	58	33.29	.00	.23	-1.20	.45

Correlation Between NCBRF Anxious and DBC Anxiety

The relationship between the DBC and NCBRF anxiety measures was investigated using a Pearson correlation measure and a similar relationship was found for both the parent and teacher data. Both were found to have a moderate strength positive correlation, whereby high anxiety on the DBC measure was associated with high anxiety on the NCBRF measure. The parent responses to the NCBRF and DBC anxiety subscales were found to be moderately correlated, $r(72) = .49$, $p < .00$. The teacher responses to the same anxiety subscales were found be similarly correlated; $r(114) = .49$, $p < .001$.

Multiple Linear Regression 1: NCBRF Anxious Parent Data

Preliminary analyses were conducted to ensure no violation of the assumptions of multicollinearity among the independent variables or with the dependent existed. The Tolerance and Variance Inflation Factor values were within acceptable parameters (Tabachnick & Fidell, 2019). Further investigations into normality, linearity, homoscedasticity, and independence of residuals were also carried out. Not all the variables met criteria for normal distribution; this limits the generalisability of these findings and will be explored further in the discussion.

A hierarchical multiple linear regression was used to predict levels of NCBRF Anxious, based on the dynamic model of autism (Bartak et al., 2006), with the outcome presented in Table 11. Steps 1 and 2 revealed non-significant models, while Step 3 revealed a significant model. DBC TPBS and NCBRF SIB at Step 3 explained 44.9% of the variance in anxiety scores, $F(10,60) = 4.90$, $p < .001$. These two measures account for an additional 24% of the variance in anxiety after controlling for the previous variables, R^2 change = .24, Fchange $(2,60) = 12.80$, $p < .001$. In the final model there were two significant variables: ID level and DBC TPBS. As ID level was reverse scored, this outcome would indicate that higher IQ scores predict higher anxiety scores on the NCBRF ($\beta = .29$, $p = .01$). Increased behaviour scores on the DBC TPBS also predicted higher anxiety scores on the NCBRF from parent data ($\beta = 54.25$, $p < .001$).

Table 11

Multiple Regression 1: The Dynamic Model of Autism, Using NCBRF Anxious–Parent Data

Variable	B	SE	β	*p*	R^2	R^2change	F	*p*
Step 1					.05	0.05	1.04	.38
Constant	17.20	13.99						
Age	.63	.48	.17	.20				
ID level	6.27	3.95	.21	.12				
SCQ	.07	.39	.02	.87				
Step 2					.22	.17	2.12	.05
Constant	-3.31	16.71						
Age	.74	.68	.20	.28				
ID level	6.51	3.85	.22	.10				
SCQ	-.27	.49	-.08	.58				
RBQ	-.38	.32	-.18	.24				
SSP2/AASP Bystander	-.49	.35	-.25	.16				
SSP2/AASP Seeker	.52	.41	.22	.21				
SSP2/AASP Avoider	.38	.38	.18	.32				
SSP2/AASP Sensor	.80	.44	.33	.07				
Step 3					.45	.24	4.90	<.001*
Constant	-5.24	14.31						

Age	1.09	.58	.29	.07
ID level	8.87	3.37	.29	.01*
SCQ	-.82	.43	-2.6	.06
RBQ	-.49	.28	-.23	.08
SSP2/AASP Bystander	-.09	.31	-.05	.77
SSP2/AASP Seeker	.07	.36	.03	.84
SSP2/AASP Avoider	.08	.33	.04	.80
SSP2/AASP Sensor	.38	.39	.16	.34
DBC TPBS	54.25	12.86	.64	<.001*
NCBRF SIB	.20	.61	.05	.74

Note. *$p < 0.01$.

Multiple Linear Regression 2: NCBRF Anxious Teacher Data

Prior to conducting the multiple linear regression, analyses were conducted to ensure no violation of the assumptions of multicollinearity among the independent variables or with the dependent existed. The Tolerance and Variance Inflation Factor values were within acceptable parameters (Tabachnick & Fidell, 2019). Investigations into normality, linearity, homoscedasticity, and independence of residuals were also carried out, with similar limitations found around the normality of the distribution within these variables, despite reasonable skewness and kurtosis statistics previously. Again, this limits the generalisability of these findings and will be explored further in the discussion.

A hierarchical multiple linear regression using the teacher NBCRF Anxious data as the dependent variable to test the dynamic model of autism (Bartak et al., 2006) was completed, and found each stage of the model to be significant. Step 1 was significant, *F*(3,110) = 5.64, $p < .001$, accounting for 13% of the variance in anxiety, with no significant variables. Step 2 was also found to be significant, *F*(8,105) = 2.61, $p = .01$, this time accounting for 17% of the variance in the model, with no significant variables. Table 12 has the outcomes.

The complete model at Step 3 accounted for 36% of the variance in NCBRF Anxious scores, *F*(10,103) = 5.82, $p < .001$. The addition of DBC TPBS and NCBRF SIB accounted for 20% of the variance in anxiety scores when the previous variables have been held constant, R^2 change = .20, *F*change (2,103) = 15.72, $p < .001$. Two variables were found to be significant in the final model: SCQ and DBC TPBS. The NCBRF teacher data found that increased SCQ scores predicted lower anxiety scores ($\beta = -.29$, $p = .003$). Increased behaviour scores on the DBC TPBS predicted higher anxiety scores ($\beta = .45$, $p < .001$).

Table 12

Multiple Regression 2: The Dynamic Model of Autism, Using NCBRF Anxiety –Teacher Data

Variable	B	SE	β	*p*	R^2	R^2change	F	*p*
Step 1					.13	.13	5.64	.001*
Constant	90.55	21.74						
Age	-1.19	.57	-.20	.04				
ID level	4.35	5.21	.09	.41				
SCQ	-1.14	.55	-.21	.04				
Step 2					.17	.03	2.61	.01*
Constant	91.09	29.41						
Age	-1.58	.76	-.27	.04				
ID level	2.50	5.39	.05	.64				
SCQ	-1.12	.58	-.21	.05				
RBQ	-.14	.10	-.14	.19				
SSP2/AASP Bystander	-.06	.34	-.02	.87				
SSP2/AASP	-.54	.46	-.01	.91				

Seeker								
SSP2/AASP Avoider	.73	.48	.18	.13				
SSP2/AASP Sensor	-.19	.65	-.04	.77				
Step 3					.36	.20	5.82	<.001*
Constant	67.37	26.34						
Age	-1.37	.69	-.23	.05				
ID level	6.35	5.04	.13	.21				
SCQ	-1.56	.52	-.29	.003*				
RBQ	-.12	.09	-.13	.17				
SSP2/AASP Bystander	.10	.09	-.13	.74				
SSP2/AASP Seeker	-.26	.41	-.07	.52				
SSP2/AASP Avoider	.56	.44	.13	.20				
SSP2/AASP Sensor	-.44	.58	-.08	.45				
DBC TPBS	59.77	14.24	.45	<.001*				
NCBRF SIB	.16	.71	.03	.82				

Note. *$p < 0.01$.

Multiple Linear Regression 3: DBC Anxiety Parent Data

Preliminary analyses were conducted to ensure no violation of the assumptions of multicollinearity among the independent variables or with the dependent existed. There was a Pearson correlation noted between DBC Anxiety and DBC TPBS of r = .79. This relationship will be explored further in the discussion, as will the impact on significant results. The Tolerance and Variance Inflation Factor values were within acceptable parameters (Tabachnick & Fidell, 2019). Investigations into normality, linearity, homoscedasticity, and independence of residuals were also carried out, with similar limitations found around the normality of the distribution within these variables, despite reasonable skewness and kurtosis statistics previously. This limits the generalisability of these findings and will be explored further in the discussion.

The hierarchical multiple linear regression was calculated to predict levels of DBC Anxiety as reported by parents, based on the dynamic model of autism (Bartak et al., 2006), with the outcome presented in Table 13. Each step of the model was found to be significant and with a different significant variable associated with explaining the variance in anxiety in Steps 1 and 3.

In Step 1, 21% of the variance in anxiety scores was accounted for by the model, $F(3,67) = 5.99$, $p = .001$. A single variable, SCQ, was found to be significant within Step 1, such that higher SCQ scores predict higher DBC Anxiety scores reported by parents, $\beta = .45$, $p < .001$. Step 2 of the Model added RBQ scores and Sensory Profile subscales, and was also found to be significant, $F(8,62) = 4.44$, $p < .001$. The Step 2 model accounted for 15% more variance in anxiety scores, which was significant, R^2 change = .15, *F*change (5,62) = 2.98, $p = .02$, with no significant individual variables.

Step 3 encompasses the full dynamic model of autism (Bartak et al., 2006), and when using DBC Anxiety with parent-recorded data as the dependent variable, the model was found to be significant and to account for 69% of the variance in anxiety scores, $F(10,60) = 13.20$, $p < .001$. A single variable, DBC TPBS, was found to be significant within the model, with increased behaviour scores predicting increased DBC Anxiety scores, ($\beta = .80$, $p < .001$). The addition of DBC TPBS and NCBRF SIB in Step 3 had a significant impact on the model, even when the previous variables are held constant. These two variables account for an additional 32% of variance within DBC anxiety, R^2 change = .32, *F*change (2,60) = 31.01, $p < .001$, with DBC TPBS as the only significant variable, playing the most significant role.

Table 13

Multiple Regression 3: The Dynamic Model of Autism, Using DBC Anxiety –Parent Data

Variable	B	SE	β	*p*	R^2	R^2change	F	*p*
Step 1					.21	.21	5.99	.001*
Constant	.10	.24						
Age	-.01	.008	-.16	.17				
ID level	.03	.07	.05	.64				
SCQ	.03	.007	.45	<.001*				
Step 2					.36	.15	4.44	<.001*
Constant	-.20	.28						
Age	-.02	.01	-.25	.13				
ID level	.02	.06	.04	.73				
SCQ	.02	.008	.29	.04				
RBQ	-.003	.005	-.09	.53				
SSP2/AASP Bystander	-.006	.006	-.17	.29				
SSP2/AASP Seeker	.009	.007	.20	.22				
SSP2/AASP Avoider	.01	.006	.31	.06				
SSP2/AASP Sensor	.008	.007	.17	.30				
Step 3					.69	.32	13.20	<.001*
Constant	-.26	.20						
Age	-.01	.008	-.14	.25				
ID level	.07	.05	.12	.18				
SCQ	.006	.006	.10	.34				
RBQ	-.005	.004	-.14	.17				
SSP2/AASP Bystander	.003	.004	.08	.50				
SSP2/AASP Seeker	.000	.005	-.01	.92				
SSP2/AASP	.006	.005	.15	.20				

Avoider				
SSP2/AASP Sensor	-.003	.006	-.06	.63
DBC TPBS	1.27	.18	.80	<.001*
NCBRF SIB	-.004	.008	-.05	.63

Note. *$p < 0.01$.

Multiple Linear Regression 4: DBC Anxiety Teacher Data

Preliminary analyses were conducted to ensure no violation of the assumptions of multicollinearity among the independent variables or with the dependent existed. The Tolerance and Variance Inflation Factor values were within acceptable parameters (Tabachnick & Fidell, 2019). Investigations into normality, linearity, homoscedasticity, and independence of residuals were also carried out, with similar limitations found around the normality of the distribution within these variables, despite reasonable skewness and kurtosis statistics previously. This limits the generalisability of these findings and will be explored further in the discussion.

A multiple linear regression was carried out using the DBC Anxiety reported by teachers as the dependent variable, with the outcome below in Table 14. Steps 2 and 3 of the model were found to be significant, with only a single variable in the final step being significant.

Step 2 was significant and accounted for 17% of the variance in anxiety scores, $F(8,106) = 2.66$, $p = .01$. However, when the additional variables are considered alone, with Step 1 held constant, their unique addition was not found to be significant. No individual variables were found to be significant.

The complete model at Step 3 was found to be significant and to account for 36% of the variance in DBC Anxiety scores as reported by teachers, $F(10,104) = 5.91$, $p < .001$. The two additional variables explained an extra 20% of the variance in anxiety scores, when controlling for the previous variables, R^2 change = .20, *F*change (2,104) = 15.92, $p < .001$. A single variable was found to be significant at this step, with increasing behaviour scores on the DBC TPBS predicting higher DBC Anxiety scores, β = .49 $p < .001$.

Table 14

Multiple Regression 4: The Dynamic Model of Autism, Using DBC Anxiety –Teacher Data

Variable	B	SE	β	*p*	R^2	R^2change	F	*p*
Step 1					.08	.08	3.25	.02
Constant	.59	.24						
Age	-.02	.006	-.23	.02				
ID level	.01	.06	.02	.86				
SCQ	.01	.006	.19	.08				
Step 2					.17	.09	2.66	.01*
Constant	-.01	.32						
Age	-.004	.008	-.06	.62				
ID level	.006	.06	.01	.92				
SCQ	.009	.006	.15	.15				
RBQ	.00	.001	.05	.66				
SSP2/AASP Bystander	-.005	.004	-.169	.20				
SSP2/AASP Seeker	.000	.005	.008	.95				
SSP2/AASP Avoider	.003	.005	.07	.52				
SSP2/AASP Sensor	.01	.007	.24	.05				
Step 3					.36	.20	5.91	<.001*
Constant	-.27	.28						
Age	-.001	.007	-.02	.88				
ID level	.04	.006	.07	.48				

SCQ	.004	.006	.07	.44
RBQ	.001	.001	.06	.53
SSP2/AASP Bystander	-.003	.003	-.10	.38
SSP2/AASP Seeker	-.002	.004	-.05	.65
SSP2/AASP Avoider	.001	.005	.02	.82
SSP2/AASP Sensor	.01	.006	.20	.06
DBC TPBS	.70	.15	.49	<.001*
NCBRF SIB	-.002	.008	-.04	.75

Note. **p* < 0.01.

Discussion

Four hierarchical multiple regressions were conducted to test the dynamic model of autism's (Bartak et al., 2006) capacity to explain the development of anxiety in individuals with ASD and ID. The model was found to be significant in each regression, regardless of the anxiety measure used or the data source, with the range in the variance of anxiety scores accounted for being 36% to 69%. Problem behaviour was consistently found to be a significant variable in each regression. Different significant variables were found to be predictive of anxiety levels based on the anxiety measure used, that is, the NCBRF or the DBC, as well as the data source (parent or teacher).

The significant variables predictive of anxiety within the model differed based on the dependent variable used, that is, NCBRF Anxious or DBC Anxiety. When NCBRF Anxious was the dependent variable, ID level and social communication as measured by the SCQ suggested that higher IQs and more social ability predicted higher anxiety scores. As the NCBRF Anxious is a more language- and performance-based measure than the DBC Anxiety, it is not surprising that it is predicted by higher IQs and lower scores on the SCQ (indicating better communication skills), as has been consistently reported in research (Davis et al., 2012; Lecavalier, 2006; Mingins et al., 2021). There are tentative data to suggest the opposite relationship with communication and the DBC Anxiety measure, with higher scores on the SCQ (indicating more communication difficulties) predicting higher anxiety in Step 1 of the parent sourced data. This is consistent with the research of Tsai (1996) and Gobrial and Raghaven (2012), but the relationship did not hold in the presence of the entire model. Both NCBRF Anxious and DBC Anxiety showed a similar relationship to DBC TPBS, whereby increasing problem behaviour predicts higher levels of anxiety on both measures. This offers an interesting inroad to considering anxiety identification from a more behavioural approach.

NCBRF Anxious

NCBRF Anxious and ID Level. As outlined previously in this chapter, the NCBRF Anxious subscale includes items focusing on language and performance anxiety, including emotional awareness and expression of that awareness. For the parent data, when the entire model was considered, decreased levels of ID (higher IQ) predicted increased levels of anxiety.

This finding supports previous research which has asserted that those with ASD and no to mild ID experience more anxiety than those with a significantly impacting ID (Gadow et al., 2005; Lecavalier, 2006; Mayes et al., 2011; Mingins et al., 2021; Muris et al., 1998; Nimmo-Smith et al., 2020; Sukhodolsky et al., 2008; Weisbrot et al., 2005). However, given that the same relationship was not found with the teacher data for the NCBRF Anxious measure or either source with the DBC Anxiety subscale, it would suggest that measurement selection plays a role in the outcome of the data and could skew the outcome significantly if the measurement tool is not well suited to the population being investigated.

NCBRF Anxious and Social Communication. The teacher data produced a significant relationship whereby, as SCQ scores increased, anxiety scores on the NCBRF were predicted to decrease. This would indicate that those with more social communication skills experience higher levels of anxiety. This supports the findings of Davis et al. (2012), Mayes et al. (2011), Kerns and Kendall (2013), and White et al. (2009). Higher scores on the SCQ are also associated with increased ASD symptomology. This would suggest that individuals with more social communication skills (and conversely, lower ASD symptoms) may be more able to express their emotions or internal experiences. Given the strong language and performance anxiety components to the NCBRF Anxious subscale, this would mean those with more social communication skills are predisposed to scoring higher on this measure than those with lower social communication scores. As such, the choice of measure could be influential in terms of the outcomes found, as the impact could be considerable.

It is also interesting that this finding from the teacher data was not replicated for the parent data set; however, this could be explained by the closer look at the questionnaire and the way it was completed by the two groups. The first question on the SCQ is, *Is s/he able to talk using short phrases or sentences?* If the respondent answers *No*, they skip ahead to Question 8; if *Yes*, they answer all the following questions, not all of which directly involve language usage. Question 1 was more commonly answered *Yes* by parents than by teachers. In looking more closely at the means of the SCQ Total, the teachers' average responses were significantly higher than those of the parents, indicating a higher level of social communication difficulty being reported. This may have been due to the larger population size available with the teachers' data, allowing for more variation in the data set. It may also be the result of parents understanding their children so well, including the use of augmented communication devices, that they consider their communication holistically rather than solely in relation to spoken language.

NCBRF Anxious and DBC TPBS. The consistent significant relationship found when testing the dynamic model of autism (Bartak et al., 2006) using the NCBRF Anxious was with the DBC TPBS as reported by both parents and teachers. DBC TPBS captures the broader notion of challenging behaviours described by Bartak et al. (2006) on the Tertiary level of difficulties that result from ongoing stress and poor intervention, causing the development of anxiety, destructiveness, physical assault, and self-injury. This finding supports previous research, which has found that increased anxiety in individuals with ASD has been associated with an increase in challenging and problematic behaviours (Moskowitz et al., 2013; Moskowitz, Walsh, et al., 2017; Rzepecka et al., 2011).

DBC Anxiety

DBC Anxiety and Social Communication. The parent data at Step 1 of the multiple regression testing the dynamic model of autism (Bartak et al., 2006) with the DBC Anxiety found a positive relationship with SCQ. This relationship is of note as it was in the opposite direction as that found with the NCBRF Anxious. For DBC Anxiety, increasing social communication difficulties predicted higher levels of anxiety. Though this finding is less widely supported, some researchers have suggested that individuals with increased communication difficulties are at increased risk of experiencing anxiety due to their communication limitations (Gobrial & Raghaven, 2012). Others have suggested that the communication difficulties result in difficulty expressing their anxiety, rather than having less likelihood of experiencing anxiety at all (Tsai, 1996). This finding, therefore, may be evidence that rather than experiencing less anxiety, individuals with significantly impacting ASD and ID have not been adequately measured or well understood, given their unique presentation. Here is a direct example of how the choice of measurement tool can impact the outcomes of research, as here it has led to contradictory findings.

DBC Anxiety and DBC TPBS. The multiple regressions testing the dynamic model of autism (Bartak et al., 2006) with the DBC Anxiety as the dependent variable with both the parent and teacher data found DBC TPBS to be a consistently significant variable. The DBC Anxiety subscale is a part of the broader DBC TPBS, and so a close relationship between the variables is not unexpected. Also of note is the large correlation noted between the DBC Anxiety and DBC TPBS within the parent data when testing for multicollinearity. As such, a strong relationship would be expected and the ability to generalise the findings limited. However, that a similar significant relationship was also found in the teacher data, where no such multicollinear relationship exists, does suggest a valid association, despite the limitations of the data and questionnaire.

This relationship indicates that increasing problem/challenging behaviour predicts an increase in anxiety. This is consistent with both the dynamic model of autism (Bartak et al., 2006) and previous research that has suggested that increased anxiety in individuals with ASD has led to an increase in problematic behaviours (Moskowitz et al., 2013; Moskowitz, Walsh, et al., 2017; Rzepecka et al., 2011). This indicates that anxiety in individuals with ASD and ID may be most evident through increasingly problematic behaviour.

The four multiple regressions were run using two different dependent variables, resulting in divergent findings particularly regarding social communication. This illustrates how measurement tool selection can significantly impact the outcome of the research being conducted and may be a considerable factor in why research to date has considered those with ASD and ID to experience meaningfully lower rates of anxiety. It may be that the measurement tools utilised in research are not appropriate to the population and therefore are not adequately capturing the internal experience of the ASD and ID population.

It is also interesting to note that while the measurement tool used was a factor for variables such as social communication and ID level, there were also some consistencies in findings, particularly with challenging behaviour, as measured by DBC TPBS. Challenging behaviours may represent a behavioural manifestation or change in presentation that is more readily available for parents and carers to note than a separate set of criteria to diagnose anxiety.

Correlation Between DBC and NCBRF

The correlations completed between the two anxiety measures used in this research reveal a clear issue: that not all anxiety measures are equal, particularly for this population. These anxiety measures were specifically chosen for their different qualities; namely, that the DBC offered a more behavioural basis, and the NCBRF a more language and internal feelings foundation. Both the parent and teacher data reported a moderate correlation between the two anxiety measures, though as has been revealed through the multiple regression, with divergent relationships to other measures.

The anxiety measures were chosen for their development within ASD/ID populations and use with research previously, as well as possibly offering different aspects to anxiety. As was found when looking directly at the items for each subscale in the Method section in this chapter, there are few direct commonalities between the subscale items of the DBC and the NCBRF. The DBC measures a small pool of behaviour-based traits; the NCBRF, while created with a disability population in mind, is clearly created for a verbal individual with access to their own internal state and the ability to share that with others. Many of the items rely on the individual's language skills and ability to access their own subjective state and communicate that to others. This may preclude these items from being easily identified as "anxiety" by parents and teachers of individuals with ASD and ID who are predominantly non-verbal and have great difficulty accessing their own internal states. This disparity between anxiety measures may account for much of the divergent research in this area. It is interesting that, as a result, quite different outcomes have been found because of these differences, which may point to a wider issue about the conclusions drawn about research for individuals with ASD and ID for whom the measurement tools are not well suited.

Limitations

This study was conducted to investigate if the dynamic model of autism (Bartak et al., 2006) offered a theoretical framework for understanding how anxiety develops in individuals with ASD and ID. The model was supported, being found to be significant in each of the four hierarchical regressions run and explaining varying proportions of the variance in anxiety; however, the findings need to be considered within the limitations of the study.

This was primarily an exploratory study, as little research, either theoretical or empirical, exists within the field. Although 52–74 participants are reasonable in research of ASD and ID (Helverschou & Martinsen, 2011; Joosten & Bundy, 2010; Joosten et al., 2012), this number was lower than ideally recommended for this type of analysis and, as such, may have limited the significant relationships that were found in the data. It also limits the generalisability into the wider ASD and ID population, and replication would be useful.

Within the data set there was also the impact of the lack of normality of distribution for several variables. Several measures were taken to address skewness and kurtosis issues when these arose more specifically, as well as accounting for residual distributions, linearity, and homoscedasticity. Nonetheless, normal distribution was not uniformly possible. A larger participant pool may have made it more likely, though the ASD and ID population is heterogenous by nature and may not comply to normal distribution expectations. The population size is nonetheless a limitation of our ability to generalise the findings from the multiple regressions beyond being an initial exploratory study.

Conclusion

This study aimed to investigate the dynamic model of autism (Bartak et al., 2006), and whether this model offered a theoretical structure for understanding the relationship between anxiety and ASD in individuals with co-occurring ID. The model was supported within this study through a series of hierarchical multiple regressions, providing an explanation for the relationship between ASD, ID, and anxiety. This relationship is clearly complex and affected by multiple factors, including the measurement tools used, particularly regarding anxiety.

In this study two measurement tools for anxiety were utilised, resulting in an apparently contradictory outcome, particularly regarding social communication, in the multiple regressions. Without understanding, acknowledging, and accounting for such issues in research, particularly within a population who cannot speak for themselves, assumptions may be made that result in a lack of understanding and in underdiagnosis, and therefore an absence of well-targeted treatment options.

Despite the divergent findings of this study, of considerable interest is the consistent outcome across all four multiple regressions that problem behaviour was found to predict increased anxiety for individuals with ASD and ID. This offers a starting point to consider a behavioural manifestation associated with anxiety, that would often be dismissed as ASD behavioural disturbance. There is research supporting ASD symptom amplification during times of mental ill health (Kerns et al., 2014; Lainhart & Folstein, 1994; Rodgers et al., 2012; Spiker et al., 2011), and research indicating the occurrence of challenging and problem behaviour associated with anxiety (Moskowitz et al., 2013; Moskowitz, Walsh, et al., 2017; Rzepecka et al., 2011). If we could start to consider behavioural changes and escalations in individuals with ASD and ID as possible signs of anxiety or mental ill health, we may have more options in how to treat and support individuals, rather than simply assuming the issues are ASD/ID based.

The dynamic model of autism (Bartak et al., 2006) is a theoretical framework which had previously not been empirically tested. In this study, it was found to provide useful insights into the relationship between ASD and anxiety for those with co-occurring ID. A key finding is how anxiety is considered and measured, with differential results found based on the measurement tool used. Given this ongoing issue considering assessment and diagnosis, and the relationship confirmed here in Study 1 between increased problem behaviour and anxiety regardless of the anxiety scale used, it would be of benefit to consider if there are broader behavioural and observable patterns associated with identifying anxiety. The behavioural manifestation of anxiety in individuals with ASD and ID will be considered in Study 2, addressed in Chapter 6.

Chapter 6: Study 2: Exploring Behavioural Indicators of Anxiety

Study 1 investigated the dynamic model of autism (Bartak et al., 2006), and found that this model offered an explanation for the development of anxiety in individuals with ASD and ID. Of particular note from this study was the consistent relationship between elevated anxiety and increased problem behaviour, regardless of the anxiety scale used or the data source. The scoping review completed in Chapter 3 highlights the numerous challenges with diagnosing anxiety in individuals with ASD and ID, such as diagnostic overshadowing, differential diagnosis, the differences between tools used to measure anxiety, and symptom amplification (Appleton et al., 2019). Given these difficulties, the high reliance on feedback from parents and carers, and the findings from Study 1, the possibility of using observable behaviours as a means of identifying anxiety is of significant interest. As such, Study 2 was designed to explore behavioural patterns and observable traits associated with anxiety in individuals with ASD and ID. Study 2 investigated the question: Are there specific behavioural patterns associated with the expression of anxiety in individuals with ASD and ID?

Study 2 utilised Bayesian model averaging (BMA), which was considered to be a particularly appropriate method of investigation given the exploratory nature of the study. As this research is exploring not only behavioural patterns but also variables of best fit into those patterns, the BMA method has the capacity to investigate models, specifically regarding a range of theoretically informed variables and their contribution to a dependent variable (Vakhitova & Alston-Knox, 2018). While a traditional linear regression will only investigate the significance of a proposed model, the aim of BMA is to investigate the merits of several models when there is less certainty about both the model itself and the possible contributions of the variables being suggested (Raftery et al., 1997). The BMA method averages the predictions of multiple plausible models rather than investigating a single significant model; this enables a more exploratory process, particularly when there is a limited population size as is the case with this study.

The population of this study is heterogeneous in nature, as was evidenced by non-compliance with a normal distribution on several variables of the data in Study 1. ASD in general is a heterogeneous condition, with diverse presentation and many possible comorbidities (Hassan & Mokhtar, 2019). The BMA process is recommended by Ranciati et al. (2018) as particularly useful when population data are heterogenous, making this technique highly relevant for the population in this study.

The BMA method was identified as the best method for this exploratory study, offering an appropriate process to investigate a specific potential behavioural phenotype for the expression of anxiety in individuals with ASD and ID. While this method is ideal when there is limited prior research for such a model, it is useful to consider previous research in the prioritising of variables when considering plausible models (Raftery, 1995). Thus, previous research was considered, including results from Study 1, in relation to the relationship between behaviour, ASD, ID, and anxiety, particularly the ways those behaviours could then be captured quantitatively.

Dependent Variable: Anxiety

As with Study 1, two anxiety measures were utilised in Study 2, as they each captured different aspects of anxiety. DBC Anxiety is behaviour based and NCBRF Anxious is performance and language based. Study 1 found these were only moderately correlated with each other and so the measures are not interchangeable. Investigation of the items in these two measures found no items in common and they appear to capture quite different aspects of anxiety. Problem behaviour was found to have a significant relationship with both anxiety measures; there were no other commonalities in the findings from Study 1 in terms of predictive variables. As anxiety continues to be the primary independent variable of concern, the two measures for anxiety—the DBC

Anxiety and NBCRF Anxious—will continue to be used, and possible behavioural patterns investigated for each measure.

Independent Variable Selection in BMA

Problem Behaviour

In Study 1, problem behaviour had a strong predictive relationship with anxiety within the dynamic model of autism (Bartak et al., 2006). This finding was consistent for both the anxiety measures and for parent and teacher data sets. Problem or challenging behaviour has also been found to be related to increases in anxiety for individuals with ASD and ID in wider research (Moskowitz et al., 2013; Moskowitz, Walsh, et al., 2017; Rzepecka et al., 2011). Problem behaviour presents a good candidate for inclusion within the BMA model.

Social Communication

Social communication was also found to be a significant variable in Study 1. In the teacher data, increased scores on the SCQ predicted lower NCBRF Anxious scores when tested within the dynamic model of autism (Bartak et al., 2006), indicating that those with more social communication skills experienced higher levels of anxiety than those with less social communication. This finding is supported by wider research, such as the work of Davis et al. (2012), Mayes et al. (2011), Kerns and Kendall (2013), and White et al. (2009), who all likewise found that individuals with ASD and high social communication abilities express higher levels of anxiety than those with reduced social communication abilities. Interestingly, this finding was not replicated when the DBC Anxiety subscale was used, which is consistent with the lack of a strong correlation between the two anxiety subscales used. It may be that the relationship between social communication and anxiety is dependent on the anxiety measurement tool used. Inclusion here will allow for further exploration. Social communication as measured by the SCQ is also indicative of ASD symptomology, with scores on the SCQ Total over

15 indicating likely ASD, with higher scores indicating more severe symptomology. There is research suggesting that ASD symptom amplification is associated with mental ill health (Lainhart & Folstein, 1994; Leyfer et al., 2006); social communication, therefore, represents a tangible option as an observable variable to be included in the BMA analysis.

Restricted and Repetitive Behaviours

While restricted and repetitive behaviours did not predict anxiety in Study 1, they have been the focus of research specifically considering anxiety and ASD (Cashin & Yorke, 2018; Davis et al., 2011; Gotham et al., 2013; Joosten et al., 2009, Rodgers et al., 2012). Rather than considering the broader concept captured by the RBQ Total score, it may be more useful to consider more specific elements of the subscales, which are more relevant when considering the current anxiety and ASD research. The four subscales available on the RBQ are Routine (adherence to routine/rigidity), Motor (repetitive motor movements), Interests (preoccupation with restricted interests), and Sensory (unusual sensory interests; Honey et al., 2012). Davis et al. (2011) discussed the high use of repetitive behaviours to reduce stress and anxiety in children with ASD when compared to controls, including both routine-based organisation, motor repetition, and echolalia.

Rodgers et al. (2012) and Gotham et al. (2013) considered *insistence on sameness* behaviours as particularly related to anxiety for those with ASD, as it allows increased control when stressed. This would be better captured by the RBQ Routine subscale, which includes items about rigidity of daily routines and need for consistency. Wigham et al. (2014) and Joyce et al. (2017) suggested that *sensory motor* behaviours may be used as an anxiety response for those with ASD. This would best equate to the RBQ Motor subscale. Subscales on the RBQ may, therefore, offer better behavioural correlates to anxiety than a total score for inclusion as variables.

Sensory Sensitivities

Sensory sensitivities or changes to the sensory profile have been found to be related to increased anxiety in those with ASD (Ben-Sassion et al., 2008; S. Green & Ben-Sasson, 2010; S. Green et al., 2012; Liss et al., 2008). Ben-Sasson et al. (2009) and Liss et al. (2008) found sensory over-responsivity (SOR) to be associated with elevated anxiety in individuals with ASD. A measure of sensory sensitivity was included in the BMA model. The Sensory Profile 2 and Adolescent/Adult Sensory Profile is divided into four domains: Avoider, Bystander, Seeker, and Sensor; SSP2/AASP Sensor was selected as a variable for inclusion in the BMA as it most closely equates with Ben-Sasson et al.'s (2009) concept of SOR.

Self-Injury

Self-injury has been linked to anxiety in individuals with ASD in prior research (Oliver et al., 2017; S. Williams et al., 2015). Self-injury increases as anxiety increases. While it was not found to be a significant variable in predicting anxiety within the dynamic model of autism (Bartak et al., 2006) in Study 1, it continues to be a variable of interest for inclusion within a model when considering behavioural manifestations of anxiety in individuals with ASD and ID and so will be included in Study 2.

Method

Setting

As with Study 1, all participants were students or clients at Giant Steps Australia, an ASD-specific preschool, school, and adult day program based in Sydney and Melbourne. An ASD-focused organisation such as Giant Steps facilitates consistency of schooling program and therapy support. Any additional therapies and interventions accessed outside of the school or adult program during the assessment period were captured via demographic information. Anxiety-related behaviours and ASD symptomology are expected to change with development. Recruitment of children and adults from across age groups provided a cross-section at various ages to allow for examination of change related to age and development.

Participants

The same set of children and young adults with an existing diagnosis of ASD and ID from Study 1 was utilised in Study 2. They were recruited through Giant Steps Australia, an independent school and adult day program specifically for individuals with a primary diagnosis of ASD. One hundred and nineteen individuals were recruited: 79 males (66.39%) and 40 females (33.61%), aged 4–27 years. The group comprised of preschool 18.50%, primary school 36.13%, high school 35.29%, and adult services 10.08%. ID level based on their diagnostic records was varied, with two individuals having no ID (1.68%), 10 having a mild ID (8.40%), 60 a moderate ID (50.42%), and 47 a severe ID (39.50%). In terms of comorbid diagnosis, 53 had an existing anxiety diagnosis (44.54%), and 47 were on daily anxiety medication (39.5%). The same group of 72 parents and 54 staff completed the questionnaires.

Materials

As with Study 1, measurement tools were chosen based on their emphasis on behavioural markers and history of use with ASD and/or ID populations. Parents/caregivers and school staff were asked to report on the presence or frequency of behaviours, rather than being asked about the underlying cause of the behaviour. See Table 15 for the questionnaires and subscales chosen following review of Study 1 and previous research; descriptions of the psychometric qualities are available in Chapter 5.

Table 15

Measurement Tools and Variables Used for Study 2

Behaviour of interest	Measurement tool	Variable
Independent variables		
Problem behaviour	*The Developmental Behaviour Checklist (DBC;* Einfeld & Tonge, 1995)	DBC TPBS
Social communication	*The Social Communication Questionnaire (SCQ;* Rutter et al., 2003)	SCQ Total
Insistence on sameness Sensory motor	*The Repetitive Behaviour Questionnaire 2 (RBQ 2;* Honey et al., 2012)	RBQ Routine (subscale) RBQ Motor (subscale)
Sensory over responsivity	*Sensory Profile 2 –Short form (SSP-2;* Dunn, 2014) *and Adolescent/Adult Sensory Profile (AASP;* Brown & Dunn, 2002)	SSP2/AASP Sensor (domain)
Self-injury	*The Nisonger Child Behaviour Rating Form (NCBRF;* Aman et al., 1996)	NCBRF SIB (subscale)
Dependent variables		
Anxiety	*The Developmental Behaviour Checklist (DBC;* Einfeld & Tonge, 1995) *The Nisonger Child Behaviour Rating Form (NCBRF;* Aman et al., 1996)	DBC Anxiety NCBRF Anxious

Procedure

All families with a child attending Giant Steps preschool, school, or adult program at either Sydney or Melbourne campuses were invited to take part in the study. Parents and caregivers were given information about the research project through parent information nights as well as through information sent home directly to families and through the weekly school newsletter. Parents were invited to participate in 2016 and then those who had not been enrolled at that time were invited in 2018, with the same information opportunities of parent information nights, the weekly school newsletter, and information sent directly home. Parents who declined to participate themselves were offered the opportunity for their child to be part of the data collection completed by staff within the school.

In 2016, 103 families were invited to take part in the research; 62 completed the questionnaires themselves (60.19%), and 99 agreed for school staff to complete further data on their children (96.12%). In 2018 an additional 36 families were invited; 10 completed the questionnaires themselves (27.78%) and 20 agreed for school staff to collect data about their children (55.56%). In total this provided data for 119 participants: 72 (52.18%) parent responses to the questionnaires (4 of which were parents only, 68 were shared responses with staff) and 115 (83.11%) staff responses for the same set of individuals with ASD and ID. Of these, 105 participants attended the Sydney school and 14 attended the Melbourne school.

Participants met the following criteria:

1. Existing clinical diagnoses of ASD and ID (with psychometric and paediatric or psychiatric reports to support this).
2. Currently enrolled at Giant Steps preschool, school, or adult program at either the Sydney or Melbourne campus.

Participants were not excluded based on level of ID or level of expressive or receptive language.

Parents were asked to complete demographic information including age, existing comorbid psychopathology, medical conditions, medication taken, schooling history, living environment (a number of students and young adults are in full- or part-time out-of-home care), family history of anxiety or other psychopathology, confirmation of ASD and ID diagnosis, and level of impairment.

For each student in the school, the questionnaires were divided amongst the professional staff working in each classroom, namely the class teacher, occupational therapist, speech therapist, and music therapist assigned to each class. Data collection took place in the second half of the year to ensure staff had a minimum of 6 months working directly with individuals and knew them well enough to answer the questions. Teachers completed the DBC and NCBRF to ensure consistency between the two anxiety rating forms. Therapists completed the SSP2/AASP, SCQ, and RBQ.

For the adults in the day program, data were completed by program staff who had worked with the individuals for a minimum of 6 months. There were no teachers and limited therapists within the day program, so support staff who worked with the clients daily were asked to complete the questionnaires.

Ethics

Ethical approval was sought and granted from both Giant Steps and the Griffith University Ethics Committee for this research to be conducted. The Griffith University Human Research Ethics reference number for this project is: 2016/010. These approvals are included in Appendices A and B.

Analysis

Given the exploratory nature of this study, each dependent variable was investigated for possible linear relationships against each of the independent variables available. This process was available through the statistical package Autostat, which was used to complete the BMA.

The process for Bayesian variable selection and estimating the most probable model relies on the specification of a prior distribution for the unknown parameters. In this model, a G-prior spike slab algorithm was used. Bayesian analysis relies on prior distributions to estimate their respective posterior distributions based on MCMC samples. In spike-slab algorithm, a variable's prior distribution is included in MCMC iterations as a mixture of two states: "slab" with a positive or negative beta coefficient, following a symmetric distribution, and "spike", a beta coefficient equal to zero. Either of these two states can be included in each of the model iterations as indicated by the stochastic search algorithm. This search is used to determine the most probable model that can explain the dependent variable, replacing the *p*-value with probability of inclusion. This probability of inclusion is based on the number of models in which the variable is included and how probable each of these models are deemed. Modelling was completed using Autostat software.

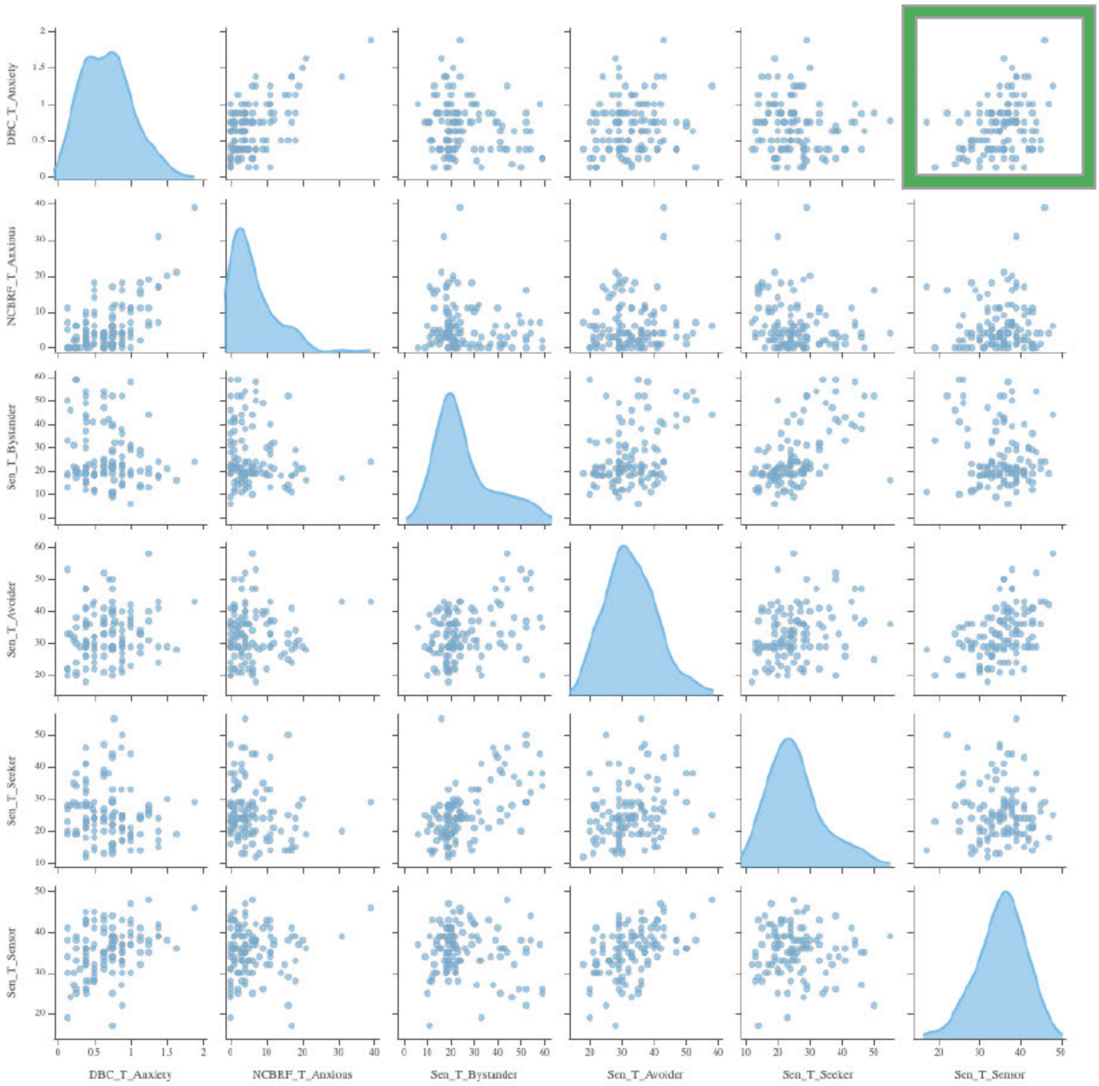
The Bayesian linear regression was conducted with the variables identified through prior research and Study 1 as being most likely to be involved in the behavioural manifestation of anxiety in individuals with ASD and ID. The following independent variables were included in the analysis (as indicated in Table 15): problem behaviour (DBC TPBS), social communication (SCQ Total), insistence on sameness repetitive behaviour (RBQ Routine), sensory motor repetitive behaviour (RBQ Motor), sensory over-responsivity (SSP2/AASP Sensor), and self-injury (NCBRF SIB). Four BMAs were completed using different dependent variables; that is, NCBRF Anxious Parent and Teacher and DBC Anxiety Parent and Teacher.

Results

The variables used in Study 2 are the same as those from Study 1. A full account of the descriptive statistics for the variables was presented in Table 10 of Chapter 5. All the variables available were investigated for possible linear relationships with anxiety, either through DBC Anxiety (Parent or Teacher) or NCBRF Anxious (Parent or Teacher). An example of this process is presented in Figure 8, which outlines how the DBC Anxiety and NCBRF Anxious teacher data relate with the four SSP2/AASP subscales: Bystander, Seeker, Avoider, and Sensor. This graph shows that the only subscale for inclusion within the model is Sensor (marked with the green square), as it offers the only possible linear relationship with DBC Anxiety. There is also a clear linear relationship between DBC Anxiety and NCBRF Anxious in these data; the two scales were confirmed in Study 1 to be moderately correlated. Following this process, no additional variables were identified for inclusion into the BMA process.

Figure 8

Distribution of DBC Anxiety and Sensory Profile Scores for Teacher Data



BMA 1: NCBRF Anxious Parent Data

A BMA was run to investigate whether the independent variables could predict the NCBRF Anxiety parent data. The BMA, in Table 16, produced the five most probable models from the six independent variables. DBC TPBS was present in each of the models, with a probability of inclusion of 1 (the highest possible score), and SCQ Total was present in four of the models, with a probability of inclusion of .93. The most probable model contained DBC TPBS and SCQ Total, with a probability of .61. The next most probable model had a significantly lower probably of .10, though it did include RBQ Motor as an additional variable. Adding either SSP2/AASP Sensor or NCBRF SIB to DBC TPBS and SCQ Total also created plausible models, but with diminishing probabilities.

Table 16

BMA 1 NCBRF Anxious Parent Probability for Inclusion Model

Variable	Inclusion probability	Model probability				
		.61	.10	.07	.06	.05
Constant	1	✓	✓	✓	✓	✓
DBC TPBS	1	✓	✓	✓	✓	✓
SCQ Total	.93	✓	✓	✓	✓	
RBQ Motor	.13		✓			
SSP2/AASP Sensor	.09			✓		
NCBRF SIB	.08				✓	
RBQ Routine	.07					

The results of the Bayesian linear regression analysis are displayed in Table 17. The DBC TPBS and SCQ Total were both found to be significant on the Bayesian linear regression, evident by the scores in the HPD columns either both being positive or negative, or one being zero. This indicates where the variable falls on the G-prior spike slab and therefore its relationship to the dependent variable. Higher scores on DBC TPBS scores predicted high anxiety scores. High DBC TPBS scores equate to more problem behaviours, both in intensity and the range of behaviours being displayed, which are therefore related to high anxiety scores on the NCBRF. Meanwhile, lower SCQ Total scores predict higher anxiety scores on the NCBRF Anxious from parent data. This means that individuals with more social communication skills, or lower ASD symptomology, but more problem behaviours have higher anxiety scores on the NCBRF Anxious according to parent data.

Table 17

Bayesian Linear Regression NCBRF Anxious Parent Data

Variable	Mean	SD	HPD 2.5%	HPD 97.5%	IF
Constant	2.04	2.33	-2.53	6.17	.94
DBC TPBS*	15.01	2.93	9.13	20.44	.99
SCQ Total*	-.29	.13	-.48	0	1.04
RBQ Motor	.03	.09	-.02	.36	1.18
SSP2/AASP Sensor	.003	.03	-.03	.05	.88
NCBRF SIB	-.002	.04	-.13	.02	.88
RBQ Routine	.006	.06	-.0002	.18	.83
Sigma	4.57	.41	3.78	5.31	1.01

Note. * significant variable on Bayesian linear regression.

BMA2: NCBRF Anxious Teacher Data

A BMA was completed using NCBRF teacher data as the dependent variable; the five most probable models are listed in Table 18. DBC TPBS and SCQ Total both have high probability of inclusions, 1 and .98 respectively. RBQ Routine had a .61 probability of inclusion, appearing in three out of the five of the probable models. The most probable model was found to include DBC TPBS, SCQ Total, and RBQ Routine and had a probability of .29. The next most probable model has these four variables, but also includes RBQ Motor, and the model probability is .21.

Table 18

BMA 2 NCBRF Anxious Teacher Probability for Inclusion

Variable	Inclusion probability	Model probability				
		.29	.21	.17	.13	.04
Constant	1	✓	✓	✓	✓	✓
DBC TPBS	1	✓	✓	✓	✓	✓
SCQ Total	.98	✓	✓	✓	✓	✓
RBQ Routine	.61	✓	✓			✓
RBQ Motor	.42		✓		✓	
NCBRF SIB	.11					✓
SSP2/AASP Sensor	.09					

The Bayesian linear regression, in Table 19, indicates four significant variables were found to be predictive of NCBRF Anxious using teacher data: DBC TPBS, SCQ Total, RBQ Routine, and RBQ Motor. This means that increasing DBC TPBS and RBQ Routine scores predicts anxiety scores will also increase, while decreasing SCQ Total and RBQ Motor scores predicts higher NCBRF Anxious scores. High RBQ Routine scores indicate increased rigidity and need for sameness, which predicts higher anxiety scores on the NCBRF Anxious. RBQ Motor scores capture repetitive motor actions such as pacing, rocking, or hand movements. This Bayesian linear regression found a negative relationship with NCBRF Anxious, meaning that decreasing RBQ Motor scores predicted higher NCBRF Anxious scores. Thus, individuals with more social communication skills, or lower ASD symptomology, increased rigidity and need for routine, reduced motor movements, and increased problem behaviour are more likely to have higher anxiety scores on the NCBRF Anxious, according to teacher data.

Table 19

Bayesian Linear Regression NCBRF Anxious Teacher Data

Variable	Mean	SD	HPD 2.5%	HPD 97.5%	IF
Constant	2.71	3.80	-4.33	10.18	1.09
DBC TPBS*	15.55	2.14	10.86	19.40	.91
SCQ Total*	-.34	.09	-.53	-.17	1.04
RBQ Routine*	.22	.21	0	.59	1.05
RBQ Motor*	-.13	.19	-.54	0	.90
NCBRF SIB	-.009	.05	-.17	.04	.95
SSP2/AASP Sensor	.002	.03	-.02	.05	.95
Sigma	5.35	.37	4.71	6.14	.98

Note. * significant variable on Bayesian linear regression.

BMA 3: DBC Anxiety Parent Data

The BMA completed using DBC Anxiety parent data, in Table 20, found two likely variables for inclusion in the most probable model: DBC TPBS and RBQ Routine. DBC TPBS appeared in all the probable models, with a probability of inclusion of 1 (the highest possible score), and RBQ Routine appeared in four out of five possible models with a probability of inclusion of .82. The model comprised of these two variables had a probability of .51. The next most probable model included only DBC TPBS and had a probably of .12.

Table 20

BMA 3 DBC Anxiety Parent Probability of Inclusion Model

Variable	Inclusion probability	Model probability				
		.51	.12	.09	.06	.05
Constant	1	✓	✓	✓	✓	✓
DBC TPBS	1	✓	✓	✓	✓	✓
RBQ Routine	.82	✓		✓	✓	✓
NCBRF SIB	.15			✓		
RBQ Motor	.11				✓	
SCQ Total	.09					✓
SSP2/AASP Sensor	.08					

The Bayesian linear regression, in Table 21, found two significant variables when looking at the HPD scores; if both these scores are positive or negative, or one is a zero, then it indicates a linear relationship with the dependent variable. This is the case for both DBC TPBS and RBQ Routine. These results indicate that increased DBC

TPBS and RBQ Routine scores predicted higher anxiety scores on the DBC. Higher scores on the RBQ Routine indicates increased rigidity and need for sameness, which together with increased problem behaviour predicts high anxiety on the DBC Anxiety.

Table 21

Bayesian Linear Regression DBC Anxiety Parent Data

Variable	Mean	SD	HPD 2.5%	HPD 97.5%	IF
Constant	-.36	.14	-.62	-.08	.95
DBC TPBS*	1.08	.16	.77	1.40	.99
RBQ Routine*	.02	.01	0	.04	.93
NCBRF SIB	-.001	.004	-.01	.001	.95
RBQ Motor	.0008	.004	-.0004	.01	.94
SCQ Total	-.0005	.002	-.007	4.68e-5	.94
SSP2/AASP Sensor	.0001	.001	-.003	.001	1.02
Sigma	.23	.02	.20	.28	.94

Note. * significant variable on Bayesian linear regression.

BMA 4: DBC Anxiety Teacher Data

A BMA was completed using DBC Anxiety teacher data as the dependent variable, shown in Table 22. The most probable model found three likely variables: RBQ Routine, DBC TPBS, and SSP2/AASP Sensor. Both RBQ Routine and DBC TPBS have a probability for inclusion of 1 (the highest score available), appearing in all five of the most probable models, while SSP2/AASP Sensor is in four of the five and has a probability of inclusion of .70. This model has a probability of .50. The next most probable model includes only RBQ Routine and DBC TPBS and has a probability of .22.

Table 22

BMA 4 DBC Anxiety Teacher Probability of Inclusion

		Model probability				
Variable	Inclusion probability	.50	.22	.08	.05	.05
Constant	1	✓	✓	✓	✓	✓
RBQ Routine	1	✓	✓	✓	✓	✓
DBC TPBS	1	✓	✓	✓	✓	✓
SSP2/AASP Sensor	.70	✓		✓	✓	✓
RBQ Motor	.13			✓		
NCBRF SIB	.10					✓
SCQ Total	.09				✓	

The Bayesian linear regression, in Table 23, found three significant independent variables: RBQ Routine, DBC TPBS, and SSP2/AASP Sensor. These results indicate that increasing scores on DBC TPBS, RBQ Routine, and SSP2/AASP Sensor all predict increasing scores on the DBC Anxiety as reported by teachers. High scores on the SSP2/AASP Sensor indicate an individual who is more likely than others to react quickly and intensely to sensory experiences. This model indicates that an individual with high problem behaviour, increased rigidity, and increased sensory sensitivity is more likely to have high anxiety on the DBC Anxiety as reported by teachers.

Table 23

Bayesian Linear Regression DBC Anxiety Teacher Data

Variable	Mean	SD	HPD 2.5%	HPD 97.5%	IF
Constant	-.52	.23	-.94	-.11	1.21
RBQ Routine*	.04	.009	.02	.05	.84
DBC TPBS*	.71	.11	.51	.93	.94
SSP2/AASP Sensor*	.008	.007	0	.02	1.09
RBQ Motor	-.0007	.004	-.01	.002	.97
NCBRF SIB	-.0003	.002	.006	.0003	.98
SCQ Total	.0001	.002	-.005	.002	.98
Sigma	.28	.02	.25	.32	.86

Note. *significant variable on Bayesian linear regression.

Discussion

This study used the BMA process to investigate behavioural patterns of anxiety in individuals with ASD and co-occurring ID. As this was a previously under-researched area, BMA offered the opportunity to create probable models from a group of variables and to consider probability of inclusion as opposed to traditional significant *p* values. Prior research, including Study 1 in Chapter 5, was considered to determine the variables to be investigated in the BMA process and six independent variables were identified.

As with Study 1, differential results were found based on the anxiety measure used. However, problem behaviour (DBC TPBS) continues to be a consistent predictive variable regardless of the anxiety measure or source of data. Another variable that was found to be predictive of anxiety in three of four BMAs completed was RBQ Routine, that is, the need for stricter adherence to schedules and insistence on sameness. SCQ Total was significant, but only with the NCBRF Anxious data, and RBQ Motor was significant for inclusion in the model with NCBRF Anxious but only with the teacher data. SSP2/AASP Sensor was found to be a significant variable only within DBC Anxiety teacher data. The two anxiety measures continue to produce different results; the BMA process has produced models of behavioural patterns associated with increased anxiety in individuals with ASD and ID. As these behavioural manifestations are more observable than many of the anxiety questionnaires or diagnostic methods currently employed, considering these behavioural changes as possible markers of anxiety offers an insight into anxiety diagnosis for this population which is otherwise challenging to diagnose.

This discussion will first consider the interpretation of the significant independent variables found through the Bayesian linear regressions for each of the anxiety measures. Then the four most probable models produced by the BMA analysis will be discussed.

Significant Independent Variables

NCBRF Anxious and DBC TPBS. Both the parent and teacher data of the NCBRF Anxious and the DBC TPBS were found to have a very high probability for inclusion across all models proposed by the BMA analysis. DBC TPBS captures a broad range of challenging and problem behaviour, including antisocial behaviour, destructiveness, self-absorbed behaviour, physical assault, and self-injury. This finding suggests that increased problem behaviour predicts higher levels of anxiety on the NCBRF Anxious scale, and supports previous research by Moskowitz et al. (2013), Moskowitz, Walsh, et al. (2017), and Rzepecka et al. (2011).

NCBRF Anxious and Social Communication. Social communication, as measured by SCQ Total, was found to be a significant predictor of NCBRF Anxious for both parent and teacher data and was included in most of the probable models offered by BMA analysis. As with Study 1, this relationship is negative; that is, as SCQ Total scores decrease, NCBRF Anxious scores increase. Higher scores on the SCQ are also associated with increased ASD symptomology. This would mean that individuals with more social communication skills and lower ASD symptoms experience higher levels of anxiety as measured by the NCBRF. These individuals are likely to have more communication skills in general and so may be able to describe their internal experience or emotions to those around them, in contrast to individuals with higher ASD symptoms and greater social communication difficulties. As mentioned in Chapter 5, the NCBRF has a strong focus on language and performance anxiety, which would predispose those with better communication skills to scoring higher on this measure. This finding supports previous research into those with ASD and anxiety, which has also found that those with more social communication skills express an increased level of anxiety (Davis et al., 2012; Kerns & Kendall, 2013; Mayes et al., 2011; White et al., 2009).

NCBRF Anxious and RBQ Routine. In the teacher data, RBQ Routine was found to be a significant predictor of anxiety, whereby as scores on RBQ Routine increase, so do scores on NCBRF Anxious. RBQ Routine measures an individual's rigidity and need for consistency on a day-to-day basis and most closely matches the concept discussed by Rodgers et al. (2012) and Gotham et al. (2013) as *insistence on sameness*. Rodgers et al. have postulated that insistence on sameness increases when anxious individuals with ASD are trying to minimise change in their environment and reduce or control their stress and anxiety. Rigidity and increased focus on routine could therefore be an observable sign of heightened anxiety in individuals with ASD and ID.

It is interesting that this finding was not replicated for the parent data. This could be because there were more teacher than parent data, allowing for trends to be more visible. Kanne et al. (2009) found that individuals with ASD responded differentially to their environment, resulting in different psychiatric symptoms. This could mean that while experiencing anxiety in both the home and school contexts, the symptoms of this anxiety may vary, rather than being identical. This could certainly explain the different results between source data.

NCBRF Anxious and RBQ Motor. RBQ Motor was found to be a significant predictor of anxiety for the teacher data. As RBQ Motor scores increase, NCBRF anxiety scores are predicted to decrease. RBQ Motor captures repetitive motor movements such as pacing, twirling, rocking, or hand mannerisms. This finding would indicate that higher anxiety scores on the NCBRF are associated with lower scores on RBQ Motor. This finding does not support Wigham et al. (2014) or Joyce et al. (2017), who suggested that increased motor movements could be used by individuals with ASD with heightened anxiety to create consistency and predictability. It is, however, more consistent with the research of Joosten et al. (2009), Joosten and Bundy (2010), and Russell et al. (2019) who suggested that sensory motor behaviours serve several purposes, of which anxiety may be but one.

In this study, the negative relationship is an interesting one, and may be due to the nature of the language and performance basis of the NCBRF Anxious scale, in that individuals scoring higher on this scale have other means of expressing their stress and anxiety such as through increased need for routine and rigidity, rather than through repetitive motor behaviours.

DBC Anxiety and DBC TPBS. The BMA analysis found the DBC TPBS had a high probability of inclusion in all the probable models and was a significant variable in terms of predicting DBC Anxiety for both parent and teacher data. The DBC Anxiety scale is a subset of the much broader DBC TPBS, so a relationship between the two variables is expected.

DBC TPBS measures a wide array of challenging behaviours of concern with a Likert scale to denote intensity of occurrence. Increasing DBC TPBS scores predict increasing anxiety scores on the DBC, which supports previous research which indicates that increased anxiety is related to increased problem behaviours in individuals with ASD (Moskowitz et al., 2013; Moskowitz, Walsh, et al., 2017; Rzepecka et al., 2011), as well as the conclusions of Study 1 in the previous chapter.

DBC Anxiety and RBQ Routine. For both the parent and teacher DBC Anxiety data, RBQ Routine was found to have a high probability of inclusion and was a significant variable in the Bayesian linear regressions. RBQ Routine most closely equates to Rodgers et al.'s (2012) concept of insistence on sameness, with the subscale aiming to capture an individual's rigidity and need for daily consistency. Increased RBQ Routine scores predicted increased DBC Anxiety. This finding is consistent with previous research by Rodgers et al. and Gotham et al. (2013).

DBC Anxiety and SSP2/AASP Sensor. The BMA analysis using the DBC Anxiety teacher data found SSP2/AASP Sensor was a significant variable in predicting anxiety. SSP2/AASP Sensor is a domain of the Sensory Profile, with high scores indicating an individual who is more likely to respond quickly and intensely to sensory experiences. SSP2/AASP Sensor was selected as it most closely resembled the concept of sensory over-responsivity, a concept discussed in research as being particularly related to anxiety (Ben-Sasson et al., 2009; Liss et al., 2008). This previous research is supported by the teacher data, in that elevated scores on the SSP2/AASP Sensor predict increased anxiety scores on the DBC.

While there are consistent findings in terms of significant variables between the anxiety measures, namely problem behaviour and insistence on sameness, there are also some notable differences. Social communication is significant with both the parent and teacher data for the NCBRF Anxious, and not at all with DBC Anxiety. This is likely due to the nature of the NCBRF Anxious scale being more language and performance based as a measure. It is not surprising that higher scores are predicted by higher communication abilities. It does suggest that the NCBRF Anxious may be better suited for those with stronger communication abilities.

The exploratory process of BMA and Bayesian linear regression was used to investigate the behavioural manifestation of anxiety in individuals with ASD and ID, using two dependent variables, the NCBRF Anxious and DBC Anxiety. Several independent variables were found to be predictive of anxiety for individuals with ASD and ID, though once again the anxiety measure and data source had a significant effect on the outcome. Problem behaviour continues to be a significant variable of interest in relation to anxiety, and insistence on sameness, as captured by RBQ Routine, was also a consistent variable across the two anxiety measures. This may suggest that while not all anxiety measures are appropriate for the ASD ID population, there are observable behaviour changes that should be considered when making a diagnosis of anxiety. Next, the four models created through the BMA process will be considered as behavioural manifestations of anxiety for individuals with ASD and ID.

Models of Behavioural Manifestation of Anxiety

The BMA analysis produced the four most probable models for the behavioural manifestation of anxiety for individuals with ASD and ID, based on the anxiety measure used and who reported the data. An overview of these models is presented in Table 24.

Table 24

Overview of Models of Behavioural Manifestation of Anxiety

Independent variable	NCBF Anxious		DBC Anxiety	
	Parent	Teacher	Parent	Teacher
Increased problem behaviour	✓	✓	✓	✓
Increased insistence on sameness		✓	✓	✓
Increased social communication	✓	✓		
Decreased repetitive motor		✓		
Increased sensory sensitivity				✓

NCBRF Anxious-Parent. The BMA analysis using NCBRF Anxious as the dependent variable and the parent data set found two significant variables in the most probable model, that is, DBC TPBS (Problem Behaviour) and SCQ Total. This indicates that the behaviours most associated with increased anxiety as measured by the NCBRF are increased challenging behaviours and increased social communication abilities or lower ASD symptomology.

NCBRF Anxious-Teacher. The BMA analysis using the teacher reported NCBRF Anxious as the dependent variable produced a model with four variables with probability for inclusion: DBC TPBS, SCQ Total, RBQ Routine, and RBQ Motor. This indicates that the behaviours most predictive of high anxiety on the NCBRF are increased problem behaviour, increased social communication abilities (lower ASD symptoms), increased need for rigidity and routine or insistence on sameness (RBQ Routine), and decreased occurrences of repetitive motor patterns such as pacing, rocking, or hand movements (RBQ Motor).

DBC Anxiety-Parent. The model produced by the BMA process using DBC Anxiety and the parent data found two significant variables, DBC TPBS and RBQ

Routine. This would indicate that increased anxiety on the DBC is predicted by increased problem behaviours and increased rigidity and need for routine, or insistence on sameness.

DBC Anxiety-Teacher. The BMA analysis using DBC Anxiety teacher as the dependent variable produced a model with three variables that were found to be most probable for inclusion: DBC TPBS, RBQ Routine, and SSP2/AASP Sensor. This would indicate that high anxiety is most related to increased challenging behaviours, increased insistence on sameness, and increased reactivity to sensory experiences.

Many of the individual relationships between the independent variables and anxiety had previously been reported in research. However, research has seldom considered a broader array of variables that together form a behavioural manifestation of anxiety within either the ASD alone or the ASD and ID combined populations. Research using cluster analysis to differentiate anxiety from other psychiatric conditions has been completed, though this has tended to focus specifically on a particular psychiatric measure, such as the work by Lecavalier (2006). This investigation aimed to assess children's behaviour according to the various subscales of the NCBRF, and how they may cluster together, creating behavioural profiles. Lecavalier used data from both parents and teachers regarding individuals with pervasive developmental delays, though not all had a formal diagnosis of ASD or ID. The parent data produced an *Anxious* profile cluster that included elevated scores on the Anxious subscale, Overly Sensitive subscale, and Conduct Problem subscale. The parent data also produced a cluster the researcher titled *Undifferentiated Behavioural Disturbance*. This cluster included even higher scores on the Anxiety subscale, as well as elevated scores on Conduct Problem, Hyperactivity, Self-Injury, Self-Isolation/Ritualistic, and Overly Sensitive, and lower scores on the Compliant and Adaptive prosocial subscales were also noted. Similar clusters were found for the teacher data; the *Anxious* cluster includes an elevated Self-

Isolation subscale score, while the *Undifferentiated Behavioural Disturbance* cluster has low scores on the prosocial subscales and significantly elevated scores on Conduct Problem, Anxious, Hyperactive, Self-Isolation/Ritualistic, and Irritable.

There are some similarities between these findings and the behaviour models found in the current research. Conduct Problems closely align with the DBC TPBS. The Self-Isolation/Ritualistic subscale is most like RBQ Motor, with more elements of personal withdrawal included, which may be why this research found an increase in ritualistic or repetitive behaviour associated with elevated anxiety. The Overly Sensitive and Irritable subscales have some overlap with the DBC Anxiety measure, but not particularly with any of the specific variables used in this research. Hyperactivity was also not looked at specifically.

The Lecavalier (2006) research has some interesting parallels in that it created empirically derived clusters to categorise behaviour profiles. The population used was not well defined in terms of diagnosis and a single questionnaire was the only information tool, resulting in limitations in terms of generalisability of the research.

Brereton et al. (2006) likewise used the DBC to assess the psychopathology of young people with ASD and ID compared to those with ID alone. Rather than using clusters based on the subscales, Brereton et al. used the behavioural subscales to describe the cohorts. The ASD cohort was defined by higher scores on all the subscales: TPBS, Anxiety, Disruptive Behaviour, Self-Absorbed Behaviour, Communication Disturbance, and Social Relating when compared to the ID alone group. This certainly supports the current research's finding of high TPBS, and Anxiety scores being related to each other. Interestingly, the increased Communication Disturbance and Social Relating (higher scores indicating increased difficulties and similar in nature to the NCBRF Self-Isolation subscale in terms of withdrawal from those around them) would indicate the opposite to the relationship found in this study between the NCBRF

Anxious and SCQ Total. Brereton et al. found that increased anxiety in those with ASD and some level of ID was related to increased communication disturbance and increased difficulties relating socially when using the DBC as both the anxiety and communication measure. The items within the Self-Absorbed subscale have strong sensory and repetitive motor components, like items on the SSP2/AASP and RBQ Motor. The Disruptive subscale does not share any items with the RBQ Routine, so does not provide a substitute for the variable Insistence on Sameness. As with Lecavalier (2006), there are limitations to considering a single information source, especially as some correlation between subscales is to be expected, particularly with TPBS, which is an overall summation of the subscales.

More recent research has also considered behavioural clustering as a means of investigating characteristics of a clinical group. J. Kim and Ha (2020) used the Child Behaviour Checklist (CBCL) to differentiate between three clusters, that is, children with ID, ASD (autistic disorder and PDD-NOS), and other mental disorders. They found that the children with ASD had significantly higher scores on all the problem behaviour syndrome subscales of the CBCL, including Anxious, Emotionally Reactive, Somatic Complaints, Withdrawn, Sleep Problems, Attention Problems, Aggressive Behaviour, Internalising, Externalising, and Total Problems. While exact correlates with the current research variables are less evident, Kim and Ha supported the idea that increased anxiety is related to multiple other behavioural issues for individuals with ASD. Again, findings are limited by the use a of single questionnaire and failure to consider how additional information could be gained from other sources that might offer valuable insights into the individual’s experience, especially for those who do not have the ability to share that experience directly.

There is a growing body of research to support the relationship between various behavioural variables and anxiety within individuals with ASD. Anxiety has been linked to other factors such as communication (Davis et al., 2011), problem behaviour (Moskowitz et al., 2013; Moskowitz, Walsh, et al., 2017), repetitive behaviours (Joosten et al., 2009), and sensory over responsivity (Ben-Sasson et al., 2008). Anxiety in individuals with ASD has also been associated with multiple behavioural disturbances and difficulties as a broader behavioural profile, including challenging behaviours, ritualistic and repetitive behaviours, and sensory sensitivities (Brereton et al., 2006; Lecavalier, 2006; J. Kim & Ha, 2020). However, scant research could be found that draws together variables from multiple sources and considers a broader behavioural phenotype that may offer more information in the complex identification of anxiety in individuals with ASD, let alone for those with ASD and ID combined.

The current research aimed to investigate behavioural patterns associated with anxiety in individuals with ASD and ID. The BMA process was used to produce four probable models based on two different anxiety measures being used and data sourced from both parents and teachers. The models produced contain both similarities and differences, probably due to the way each questionnaire measures anxiety. The NCBRF Anxious is a language- and performance-based anxiety measure, with high anxiety scores predicted by increased challenging behaviours and increased social communication skills (and therefore decreased ASD symptomology). The teacher data also noted increased insistence in sameness and decreased incidence of repetitive motor actions. Increased scores on the DBC Anxiety were predicted by increased challenging behaviours and increased insistence on sameness, with the teacher data additionally noting increased sensory sensitivity. The similarities between the models confirm there is a potential behaviour profile associated with anxiety for this population, regardless of the measure used, with challenging behaviour and insistence on sameness being strong contenders for inclusion within that model. These factors were significant with both anxiety measures despite them only having a moderate correlation in the study reported in Chapter 5.

The differences between the models could be due to the measurement tools themselves. The NCBRF Anxious focuses on language- and performance-based anxiety, while DBC Anxiety is behaviourally based. This would indicate that the NCBRF Anxious, and others like it, are better suited for use with individuals with more communication abilities and may not be appropriate for use with those who are predominantly non-verbal. As such, the DBC Anxiety may offer a more accurate measure of anxiety for all the participants in this study, exclusive of their communication abilities or social relating skills. Choice of anxiety measurement tool is therefore a significant factor to consider in both the interpretation of research and in future planning, as the impact of the tool on outcomes may be considerable.

Study 2 investigated behavioural patterns of anxiety in individuals with ASD and co-occurring ID. Four probable models were produced, with behavioural patterns associated with increased anxiety scores on two different measures. The consistencies in models between the two measures offer an insight into observable traits associated with elevated anxiety in individuals with ASD and ID. The differences in models indicate the ongoing concerns of appropriateness of measure selection for the population under investigation.

Limitations

This was an exploratory study, with limited research considering behavioural profiles or models of anxiety either theoretically or empirically. Previous research was used for variable selection and inclusion in the study; however, there is a lack of prior research for the broader considerations of behavioural models within the ASD and ID population. As such, the findings of this research are a starting point for further research, as they offer an interesting potential for increased understanding of individuals with ASD and ID.

As with Study 1, population size is a limitation, particularly when considering the parent data sources. Although 52–74 participants are reasonable in research of ASD and ID (Helverschou & Martinsen, 2011; Joosten & Bundy, 2010; Joosten et al., 2012), the parent size for this analysis was 72 and for teachers was 115. While not a small population size, given the number of variables used within the analysis, there is a limit to the power and generalisability of the results, particularly given the highly heterogenous nature of the population being investigated. As such, this initial exploratory study should be replicated.

Conclusion

This study aimed to investigate the behavioural patterns associated with anxiety in individuals with ASD and ID. The BMA process produced the four most probable models associated with parent and teacher reported data for each anxiety measure. While problem behaviour and insistence on sameness were generally significant across the anxiety measures, there were other variables that were significant with a specific measure and data source, such as social communication, repetitive motor actions, and sensory sensitivities. As was found in Study 1, the measurement tool used has played a significant role in the findings in relation to this complex relationship between behaviour, anxiety, ASD, and ID.

Limited previous research could be found that considered behavioural patterns associated with anxiety for individuals with ASD, let alone for those with co-occurring ID, or that included multiple information sources. This exploratory research offers a prospective conceptualisation for identification of anxiety in those with ASD and ID.

Chapter 7: General Discussion

In this chapter, the findings from the two studies conducted as part of this research are drawn together and considered in the context of previous research. The limitations of the current research and future directions are discussed. The contribution of this research to the broader field is also established. The aim of this thesis was to synthesise available research, considering a range of sources, particularly given the lack of specific inquiry into this specific population. A scoping review of existing research has been produced, consideration given as to the theoretical underpinnings as to the development of anxiety for individuals with ASD and ID, and two studies completed to address the following questions:

1. Does the dynamic model of autism explain the relationship between anxiety and ASD for individuals with co-occurring ID?
2. Are there specific behavioural patterns for the expression of anxiety in individuals with ASD and ID?

These questions were considered particularly pertinent given the lack of previous research into this population, as outlined in the scoping review (Chapter 3) identifying the difficulties of diagnosing anxiety in those with ASD and ID (Appleton et al., 2019). After exploring the theories of anxiety (Chapter 4), the dynamic model of autism (Bartak et al., 2006) was identified as offering a theoretical structure for understanding the development of anxiety in those with ASD, without being exclusionary on the basis of language, comprehension, or ID level. Results from Study 1 (Chapter 5) suggest that the dynamic model of autism does offer an explanation for how anxiety develops in those with ASD and co-occurring ID, although this requires further validation. Behavioural models were produced in Study 2 (Chapter 6), and results suggested increased problem behaviour and insistence on sameness behaviour were most likely to predict increased anxiety, regardless of the anxiety measure used or the data source.

Anxiety is a prevalent issue for individuals with ASD, although there is currently limited research into how it presents for those with ASD and co-occurring ID (Appleton et al., 2019). In Chapter 4, multiple theoretical frameworks for the development of anxiety were considered, including biological, physiological, learning- and attention-based, and ASD-specific theories. A key limitation of many of these theoretical frameworks was the assumption of participants' cognitive or language abilities, which effectively excluded those with a comorbid ID. Only one theory, the dynamic model of autism (Bartak et al., 2006), was found that could be applied to those with co-occurring ASD and ID. This theory offered the most plausible framework for considering how anxiety is related to ASD in those with co-occurring ID.

The difficulties identified in diagnosing anxiety in individuals with ASD and ID are likely to lead to under-reporting, limited and/or ineffective treatment options, and scant research into this specific population (Appleton et al., 2019). A review of recent research, in Chapter 3, noted issues such as diagnostic overshadowing, differential diagnosis, symptom amplification, and difficulties with measurement tools as key limitations of the current diagnostic process. Behavioural patterns associated with anxiety may be potential diagnostic indicators for those with ASD and ID and may offer an alternative method of identification.

In this chapter the key research questions are reviewed individually and then together. Their relevance to previous research is considered before the implications for clinical work and future research are explored.

Study 1: The Dynamic Model of Autism

In Study 1, the capacity for the dynamic model of autism (Bartak et al., 2006) to explain the relationship between anxiety and ASD in those with co-occurring ID was addressed. Four hierarchical multiple regressions were computed, with parent and teacher data sets and two different anxiety measures used as dependent variables, namely the NCRBF Anxious and the DBC Anxiety. Each of these regressions was found to be significant, suggesting that the dynamic model of autism does indeed explain how anxiety develops in those with ASD and ID, with the independent variable, problem behaviour, being a consistent predictor of anxiety in each regression. However, there were differences between parent and teacher reporting and between the anxiety measures, resulting in variances in additional significant independent variables that were only significant within a single regression, namely, ID level and social communication. Variance was found between the multiple regressions as to whether each step of the model was significant, or only the model in its entirety. For example, the NCBRF Anxious as reported by parents found that ID level as well as problem behaviour were predictive of anxiety, and only the entire model was significant, with Steps 1 and 2 not being significant.

The key findings will now be considered from Study 1; that is, the patterns of significance within the multiple regressions for both parent and teacher data, then the independent contributing factors found to be predictive of anxiety will be addressed, specifically problem behaviour, ID level, and social communication; the lack of further contributing factors to the model found in the data will then be considered.

Patterns of Significance in the Dynamic Model of Autism

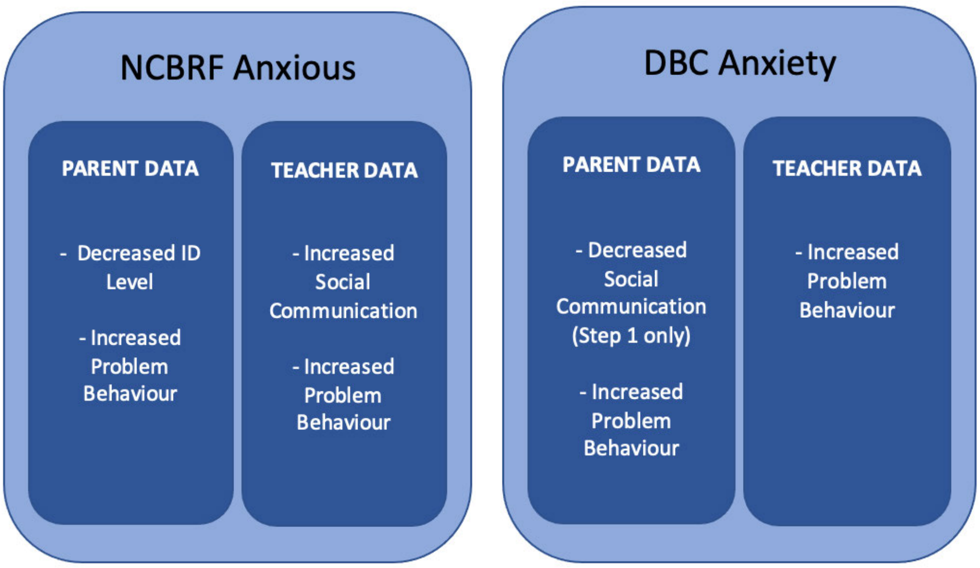
The multiple regression using NCBRF Anxious parent data to investigate the dynamic model of autism (Bartak et al., 2006) was significant when the entire model was considered, with problem behaviour and ID level both being contributing factors predictive of anxiety at the final step. However, the first two steps of the model were not found to be significant, nor were any independent variables found to be significant in Steps 1 or 2. The regression using NCBRF Anxious teacher data was found to be significant at each step of the analysis, including when the model was considered as a whole. Problem behaviour and social communication were found to be contributing factors at the final stage of the model. SCQ Total was a significant contributing variable within the NCBRF Anxious teacher data when the entire model is taken into account, with decreasing SCQ Total score predicting increasing anxiety scores. This would indicate that individuals with more social communication abilities experience more anxiety. This is consistent with previous research that has likewise found that individuals with increased social communication report higher levels of anxiety (Davis et al., 2012; Kerns & Kendall, 2013; Mayes et al., 2011; White et al., 2009).

The regression using DBC Anxiety teacher data found significant results for Step 2 and the entire model, but Step 1 was not significant. Problem behaviour was the only independent significant contributing factor that was predictive of anxiety and this was in the final step of the model. However, the parent data from the DBC Anxiety were found to be significant at each step of the analysis, including when the model was considered as a whole. Problem behaviour, as with each model, was a significant independent contributing variable, but interestingly the role of SCQ Total seems to be somewhat contradictory when compared to the NCBRF Anxious teacher results. The DBC Anxiety parent data (with only Age, ID level, and SCQ Total entered into the model as variables predicting anxiety) found at Step 1 that SCQ Total was a significant contributing factor. With DBC Anxiety, increased SCQ Total scores were predictive of increased anxiety, indicating that individuals with decreased social communication abilities experienced increased anxiety when limited other variables (Age, ID level, and SCQ Total) were considered. This finding is less commonly reported in research, although it has been noted (Gobrial & Raghaven, 2012; Tsai, 1996). The relationship does not remain significant when the entire model is analysed, indicating that other variables overshadow the relationship between SCQ Total and DBC Anxiety. However, the opposing direction noted between SCQ Total and the two anxiety measures is of note as it indicates the effect that the anxiety measurement tool choice potentially has on the outcome. This issue will be considered further when the anxiety measures are examined more closely later in this chapter. In Figure 9, an overview of the independent contributing factors for each anxiety measure and data source is shown.

It is therefore unclear how much each step within the dynamic model of autism (Bartak et al., 2006) contributes to the model overall, as not every step was found to be significant. There is also a lack of significant contributing variables found at each step of the model to account for additional anxiety. This issue will be considered in depth later in the chapter, particularly in relation to support for the hierarchical nature of the model, including the question of whether we have adequate means of measuring each independent variable for this complex population. Nevertheless, when considered as a whole and regardless of anxiety measurement tool or data source, the dynamic model of autism was found to explain the development of anxiety in individuals with ASD and ID.

Figure 9

Overview of Independent Contributing Variables from Study 1



Independent Contributing Factor: Problem Behaviour

Problem behaviour was consistently found to be a significant contributing factor in predicting anxiety in each of the hierarchical multiple regressions completed, regardless of anxiety measurement tool or data source. This indicated that increased problem behaviours predicted increased anxiety on both the NCBRF Anxious and DBC Anxiety for parent and teacher data. This finding is consistent with previous research that likewise found problem behaviour increased with anxiety in those with ASD and ID (Moskowitz et al., 2013; Moskowitz, Walsh, et al., 2017; Rzepecka et al., 2011). Moskowitz et al. (2013) found that problem behaviour was significantly related to increased measures of physiological stress (including changes in heart rate and respiratory sinus arrhythmia), and was also responsive to anxiety treatment protocols, reducing when supported through ASD altered cognitive behavioural therapy treatment (Moskowitz, Walsh, et al., 2017). Moskowitz, Rosen, et al. (2017) also noted in their recommendations around multimodal assessment for anxiety in those with ASD that including measures of problem behaviour could be an additional diagnostic feature, particularly for those with minimal language and reduced ability to communicate their internal experience.

While there is limited research into the affiliation between anxiety and problem behaviour for individuals with ASD and ID, there is research into this relationship within the ASD and ID populations separately. Bitsika and Sharpley (2016) investigated children with ASD and no ID and found that irritability and temper tantrums were positively associated with increased anxiety. Koritsas and Iacono (2015) found that challenging behaviours were predicted by anxiety scores and severity of ID, indicating that those with increased anxiety and severe to profound level ID showed more challenging behaviours than their mild to moderate ID and lower anxiety counterparts. A literature review of anxiety and challenging behaviour in individuals with ID reported a consistent relationship between anxiety and challenging behaviours, whereby as one increases the other appears to as well, though the exact nature of the association was unclear (Pruijssers et al., 2014). There is broader research supporting this relationship between problem or challenging behaviour and anxiety in individuals with ASD or ID, and those with co-occurring ASD and ID. It was certainly found to be consistently significant in the current research, and together with prior research, is of interest when considering the behavioural manifestation of anxiety in individuals with ASD and ID.

Independent Contributing Factor: ID Level

When testing the dynamic model of autism (Bartak et al., 2006), the parent data found level of ID to be a significant contributing factor in predicting increased NCBRF Anxious scores. This indicated that higher levels of anxiety, as recorded by the NCBRF, were experienced by those with higher IQs, a finding which is consistent with previous research, particularly in children with ASD (Gadow et al., 2005; Lecavalier, 2006; Mayes et al., 2011; Mingins et al., 2021; Muris et al., 1998; Nimmo-Smith et al., 2020; Sukhodolsky et al., 2008; Weisbrot et al., 2005). While the finding in the current study is consistent with prior research, it is of note that this finding was not replicated within the teacher data set for the NCBRF Anxious dynamic model of autism regression. Differences between parent and teacher reports for ASD populations is a consistent finding in research particularly when investigating anxiety (Adams et al., 2018; Adams et al., 2019; Gadow et al., 2004, 2006). Kanne et al. (2009) have postulated that this difference is specific to ASD populations and that inter-rater agreement between parents and teachers for students is often more reliable for typically developing children. They argued that psychiatric symptom expression varies across environments for individuals with ASD, and so rather than being a lack of understanding from either the parents or teachers, it may actually represent a different presentation of the individual in a different context. This change may account for some of the variance found in the data here, with the individuals in the study responding or presenting differently at home and at school.

Independent Contributing Factor: Social Communication

A second significant independent variable was found within the NCBRF Anxious teacher data set, with decreased SCQ Total scores predicting increased anxiety. As the social communication measure is also indicative of ASD symptomology, this would suggest that individuals with more social communication abilities or less ASD symptoms experience higher levels of anxiety. This finding is supported by previous research, which has likewise found that individuals who have ASD and who have the ability to share their experiences, report increased levels of anxiety (Davis et al., 2012; Kerns & Kendall, 2013; Mayes et al., 2011; White et al., 2009).

As the SCQ Total can also be used as a broad indicator of ASD symptomology, it is also useful to consider where this finding sits within more general ASD symptom research in terms of anxiety. Hunsche et al. (2022) noted the link between social communication and social functioning, particularly when considering the occurrence of anxiety. They noted that social functioning decreases in individuals with ASD as anxiety increases. This finding has been supported by other research (Bishop-Fitzpatrick et al., 2013; Chang et al., 2012). These findings are contrary to the current research, as they suggest that anxiety increases as ASD symptoms also increase.

This difference in findings could be explained by considering the structure of the NCBRF Anxious scale itself. The NCBRF Anxious focuses on language and performance anxiety, meaning that those with increased language or communication skills are predisposed to score higher on the measure than those with less social communication and language abilities. As neither ID/IQ or social communication were found to be predictive of high levels of DBC Anxiety, it appears the choice of anxiety measure can impact the outcome of the data and could potentially influence the conclusions being drawn.

The parent data do not replicate the teacher data. As was mentioned above, this may indicate that the individuals in the study present differently across the environments, with their communication in particular presenting differently between the home and school contexts (Kanne et al., 2009).

Missing Independent Contributing Factors: Sensory and Repetitive Behaviours

In testing the dynamic model of autism (Bartak et al., 2006), the four hierarchical multiple regressions were found to be significant and three significant independent contributing factors were found to be predictive of increased anxiety. Of particular interest within any of the four analyses was the lack of significant contributing factors within the second step of the model, specifically, sensory issues or repetitive behaviours. The lack of significance for these variables raises the question as to whether the dynamic model of autism's Step 2 is a necessary stage in its hierarchy, or alternatively, if the measurement tools available are able to accurately capture the necessary data. This will now be explored.

Sensory responsivity issues are a key component of the secondary level of the dynamic model of autism (Bartak et al., 2006). In Study 1, sensory issues were not found to be a significant predictive factor in any of the multiple regressions completed. However, there is research to support a relationship between sensory integration issues or sensory over-responsivity and heightened anxiety in children with ASD (Ben-Sasson et al., 2008; Black et al., 2017; S. Green et al., 2012; South & Rodgers, 2017). Syu et al. (2020) also found a similar relationship between sensory over-responsivity and increased anxiety in adults with ASD.

As no significant relationship was found in this study between sensory sensitivity and anxiety, it is interesting to consider why this may be the case. To understand this, a closer look must be taken at the questionnaire used to measure sensory integration issues. In this study, two forms of the Sensory Profile were used to gather information to accommodate the wide age range of the participants: The Short Sensory Profile 2 and the Adolescent/Adult Sensory Profile. While both questionnaires produce the same four quadrant outcomes of Avoider, Bystander, Seeker, and Sensor, the fact that there has been an update to the test items in the SSP2, but not in the AASP, may be a factor in this study. The AASP is divided up more specifically into sensory experiences, covering taste, touch, smell, visual, auditory, movement, and activity levels. Responses in relation to how much the individual seeks, avoids, struggles with, or may be distracted by these various experiences are then reassessed into the quadrants and compared to typically developing data to create their profile. By comparison, the items within the SSP2 were changed significantly, focusing on *Sensory Processing* with items such as "moves stiffly" and "rocks in chair" and *Behavioural Responses to Sensory Processing* with items such as "gets frustrated easily" and "struggles to pay attention", rather than sensory experiences themselves, yet still producing the same quadrant outcomes. As a result, while all the participants' sensory data are analysed together, there may be a lack of consistency between the data collected from the SSP2 for those aged 4–14 years old and from the AASP for those aged +14 years. Despite this possibility, the SSP2 is a widely used sensory measure in research, with Burns et al. (2017) reporting in their systematic review of sensory assessments in ASD that 79% of research papers used the SP or SP2. They also reported that the SP/SP2 was appropriate to use with those with ASD, despite not being created for a disability population. While little previous research could be found that had used both the SSP2 and AASP with an ASD population due mostly to the ages of cohorts being studied, Kolacz et al. (2018) did use a combination of sensory profile questionnaires and found significant results with an ASD population. However, their research was primarily interested in those with co-occurring Fragile-X and the level of cognitive functioning of the participants was not mentioned, so it is unclear what role ID may be playing in interpreting these results.

Another key component in the secondary level of the dynamic model of autism (Bartak et al., 2006) is repetitive or ritualistic behaviours. This was not found to be a significant contributing factor in predicting anxiety in any of the four regressions completed. Despite this, prior research has found that increased repetitive behaviours in individuals with ASD have been associated with heightened anxiety (Cashin & Yorke, 2018; Gotham et al., 2013; Joosten et al., 2009; Rodgers et al., 2012). Within Study 1, repetitive behaviours were considered as a whole, and measured using RBQ Total, representing the combination of the four subscales: Routines, Motor, Interests, and Sensory. It may therefore be the case that a stronger and more predictive relationship with anxiety within the dynamic model of autism may have been found if the discrete characteristics of repetitive and ritualistic behaviours were considered, rather than the more general factors.

A Summary of Support for the Dynamic Model of Autism

The dynamic model of autism (Bartak et al., 2006) was supported by each of the multiple regressions completed. This suggests that the model explains why individuals with ASD are particularly susceptible to developing anxiety, as the very nature of their diagnosis and the way they process information makes them vulnerable to experiencing stress which, if not adequately supported or addressed, can in turn lead to the diagnosis of an anxiety disorder. It also points to the role that carers, professionals, and support staff play in potentially removing the coping strategies individuals employ to manage environmental stressors (Bartak et al., 2006). Importantly, the dynamic model of autism does not exclude those with an ID in its explanation for how anxiety develops in those with ASD. In fact, individuals with ASD and ID may be expected to experience higher levels of anxiety than those with ASD alone, when considering the impact of language and information processing deficits at each level of the dynamic model of autism.

The dynamic model of autism (Bartak et al., 2006) was found to be significant for the parent and teacher data sets when NCBRF Anxious was the dependent variable. Increased NCBRF Anxious scores were predicted by decreased ID scores (increased IQ) on the parent data or increased social communication abilities for the teacher data. It is worth considering what this means within the broader context of the dynamic model of autism. In their model, Bartak et al. included communication and information processing deficits in the primary level of difficulties, indicating that the presence of these deficits interacting with environmental stressors leads to secondary impacts of ASD, such as sensory modulation issues, odd social behaviours, and insistence on sameness and repetitive behaviours as coping mechanisms. The authors stated that skill development to mitigate these primary level deficits can lead to secondary level gains, such as increased sensory modulation ability as language and social skills improve. In this way, treatment and intervention is indicated at various points of the model, and by addressing different aspects of behaviour, multiple impacts on different levels may be possible over time. As such, individuals with ASD who have increased social communication, language, and/or higher IQ could also have the ability to learn new coping skills and strategies more readily than their counterparts with a co-occurring ID. This could have a considerable flow-on effect on their secondary and, ultimately, tertiary and quaternary level behaviours. In fact, individuals with ASD and fewer social communication skills, less language, or more co-occurring ID could be seen in the dynamic model of autism to be even more susceptible to the impact of stress and developing anxiety, while also having less opportunity to develop skills and alternative coping strategies to change the trajectory of their experience.

Analyses using DBC Anxiety as the dependent variable were also significant, and showed that the only significant independent variable was problem behaviour (which was significant in each analysis). Within this analysis neither IQ nor social communication were found to be significant variables. The overall findings are more consistent with the narrative of anxiety development provided by the dynamic model of autism (Bartak et al., 2006).

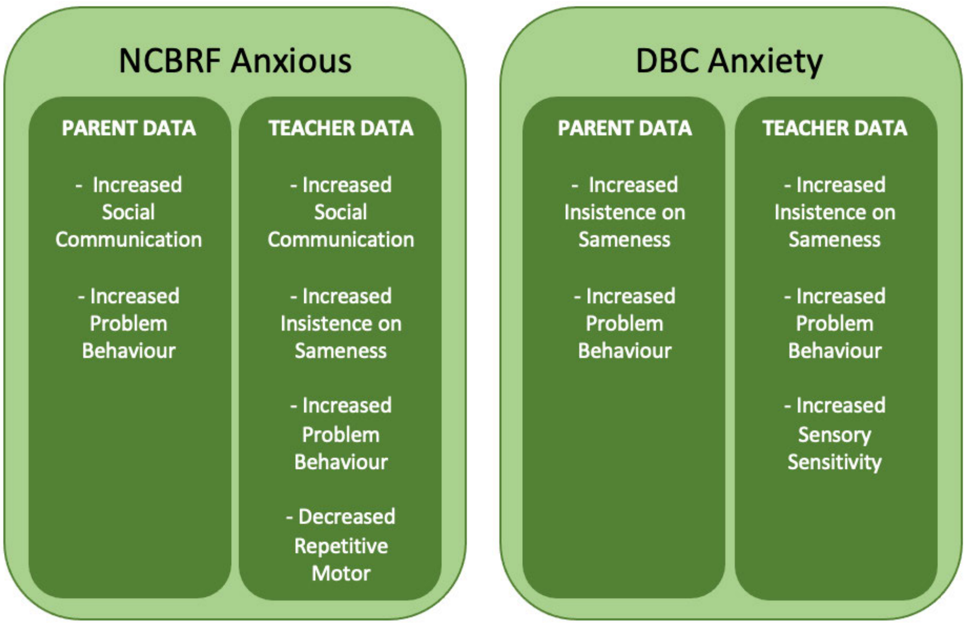
In conclusion, the aim of Study 1 was to investigate whether the dynamic model of autism (Bartak et al., 2006) explained the development of anxiety in individuals with ASD and co-occurring ID. The model was tested using hierarchical multiple regressions, and was found to be significant, indicating it does explain the development of anxiety within this under-researched population. The model was significant across both the anxiety measures used, despite the differences between those measures, adding to the veracity of the model itself for its use across the autism spectrum and with individuals with and without an ID. On closer inspection of the significant independent variables, of particular interest was problem behaviour, which was significant in all four multiple regressions in predicting increased anxiety. This offers a starting point to consider a behavioural manifestation associated with anxiety, that has been shown to be identifiable by carers but that may often be dismissed as ASD behavioural disturbance. If we could start to consider behavioural changes and escalations in individuals with ASD and ID as possible signs of anxiety or mental ill health, we may have more options in how to treat and support individuals, rather than simply assuming the issues are ASD/ID based. This warrants further research. Further consideration of problem behaviour as a marker of anxiety in this under-researched population will be considered in the discussion of Study 2.

Study 2: Behavioural Manifestation of Anxiety

There are numerous challenges associated with diagnosing anxiety in those with ASD and ID (Appleton et al., 2019). The identification of behavioural patterns accompanying anxiety in these individuals offers a potentially more reliable form of identification. In Study 1, problem behaviour was consistently found to predict increased anxiety regardless of the data source or anxiety measure used. It therefore represents an observable trait that could be utilised to identify anxiety in individuals with ASD and ID. Study 2 investigated the research question: Are there specific behavioural patterns associated with the expression of anxiety in individuals with ASD and ID? using BMA to create probable models. The individual significant independent variables found (see Figure 10) are reviewed and then the most probable models considered in light of the dynamic model of autism (Bartak et al., 2006). Clinical implications, the limitations associated with the current research, and future research and are then discussed.

Figure 10

Overview of Significant Independent Variables in Study 2



Significant Independent Variables

Problem Behaviour. Across the DBC Anxiety and NCBRF Anxious, and for both parent and teacher reported data, increasing problem behaviour was found to be predictive of increasing anxiety scores. Problem behaviour was measured by the DBC TPBS and included a broad range of behaviours of concern such as antisocial behaviour, destructiveness, self-absorbed behaviour, physical assault, and self-injury. This finding is consistent with prior research, which likewise found that problem behaviour increased during times of heightened anxiety for individuals with ASD and ID (Moskowitz et al., 2013; Rzepecka et al., 2011). Problem behaviour therefore seems to be a plausible marker of increased anxiety in individuals with ASD and ID, particularly as it is identifiable by both parents and teachers.

Insistence on Sameness. For three out of four of the most probable models produced from the BMA process, RBQ Routine was found to be a significant variable in predicting anxiety. This was the case for the NCBRF Anxious teacher data and for both teacher and parent data for DBC Anxiety. For each of these, increased scores on RBQ Routine predicted increased anxiety scores. RBQ Routine captures an individual's rigidity, need for consistency, and adherence to daily routines. This subscale of the RBQ Total was chosen for its close resemblance to the concept discussed by Rodgers et al. (2012) and Gotham et al. (2013) as *insistence on sameness.* Rodgers et al. and Gotham et al. likewise used the RBQ in their research. These findings support the prior research of Rodgers et al., who suggested that insistence on sameness is similar in concept to *intolerance of uncertainty* within the well-established mainstream model of anxiety created by Dugas et al. (1998). Intolerance of uncertainty assumes that uncertainty is stressful and that unpredictable events are negative in nature and therefore to be avoided. Rodgers et al. postulated that this is very similar to insistence on sameness for those with ASD, in that there is a need for consistency and predictability to avoid undue stress and anxiety.

Routines or insistence on sameness may ameliorate anxiety in the short-term by reducing immediate demands and restricting the environment. Rodgers et al. (2012) theorised that the immediate anxiety benefits of insistence of sameness behaviours results in positive beliefs about the function of these behaviours, further imbedding their use and maintaining them into the longer term. This decreases the likelihood of individuals with ASD finding other strategies for managing stress or engaging with the wider environment, thus perpetuating the anxiety cycle. This is based on Dugas et al.'s (1998) anxiety theory, which is highly cognitive in nature and conceptualised for a mainstream population (as discussed in Chapter 4). Dugas et al.'s anxiety theory was not constructed for an ASD population, and with its cognitive focus it excludes those within the current study. Yet, the postulated theory by Rodgers et al. of insistence on sameness or intolerance of uncertainty being related to increased anxiety has been supported in this study, regardless of the presence of significant cognitive difficulties within the population being studied. The findings of this research are also supported by the work of Gotham et al. (2013), who likewise found that insistence on sameness, unlike other measures of anxiety such as language-based items, was unrelated to IQ or level of ID. Insistence on sameness, as measured by increased need for routine and consistency, therefore requires further investigation, as it appears to be a relevant factor in the experience of anxiety for individuals with ASD, including those with co-occurring ID.

This finding was consistent with both the DBC Anxiety and NCBRF Anxious, although only the teacher data were found to be significant in the NCBRF Anxious data. This may be due in part to the disparities between the measures themselves, in addition to the differences between the environments being reported upon. As has been previously discussed, the NCBRF Anxious scale may be more applicable for use with individuals with higher social communication abilities, as items rely more heavily on language ability and performance anxiety. The DBC Anxiety is more behaviourally based and does not have the same relationship with social communication. Kanne et al. (2009) noted that psychiatric symptomology may differ between environments as a result of the individual with ASD responding differentially to the context. As such, stress or anxiety may look different for the same individual in different environments.

Insistence on sameness therefore seems to have significant potential as a behavioural marker for anxiety. It has been shown to be identifiable by both parents and school staff, and consistently found to be predictive of increased anxiety across both measurement tools. Further research into insistence on sameness is certainly warranted.

Social Communication. Models created with the parent and teacher data found that increased scores on the SCQ Total predicted decreased scores on the NCBRF Anxious outcome measure. This indicates that those with more social communication abilities (and lower ASD symptomology) are likely to express higher levels of anxiety as measured by the NCBRF Anxious subscale. This finding is consistent with the outcome of Study 1, which likewise found that higher anxiety on the NCBRF Anxious was predicted by higher social communication abilities. This is also consistent with wider research that has found that those with more communication skills express higher levels of anxiety (Davis et al., 2012; Kerns & Kendall, 2013; Mayes et al., 2011; White et al., 2009). As with Study 1, it is inconsistent with research that suggest that increased ASD symptoms, such as increased difficulties with social functioning and theory of mind impairments, are related to increased anxiety in children with ASD (Bishop-Fitzpatrick et al., 2013; Chang et al., 2012; Hunsche et al., 2022).

This finding was not replicated with the DBC Anxiety measure, suggesting that this is not a universal relationship, but instead one that is measurement tool specific. As has been discussed above in relation to Study 1, there are significant differences between the items of the NCBRF Anxious and DBC Anxiety subscales, despite both claiming to measure anxiety. This difference in content was also noted by Reardon et al. (2015) in their review of anxiety assessment tools for children with ID. The authors particularly noted the lack of overlap in the content between these two measurement tools, with the DBC focusing on distress and behavioural factors and the NCBRF on sensitivity and desirability/performance items. When the correlation of the two measures was run, a moderate strength correlation was found. However, research using a non-ASD population investigating the factor analysis of the NCBRF and comparing it directly to the DBC reported only a small correlation between the anxiety subscales of $r = .39$ (Norris & Lecavalier, 2011). It may therefore be the case that not all anxiety measures are appropriate for use with individuals with ASD and ID, particularly those that rely more heavily on language, performance, or cognition-based items for anxiety. Those with ASD and ID are unlikely to have the communication skills to describe their experience of anxiety. This discrepancy is evident in the differences found between the two anxiety measures within the current research, even though both were chosen because they were designed for a disability population. As a result of this inconsistent relationship with anxiety, social communication does not represent a good behavioural marker for identifying or diagnosing anxiety in individuals with ASD and co-occurring ID.

Sensory Sensitivity. The Bayesian model created with the teacher-reported data found that increased sensory sensitivity predicted increased anxiety as measured by the DBC Anxiety. Sensory sensitivity was measured by the Short Sensory Profile 2 and Adolescent/Adult Sensory Profile, which both produced four quadrants: Seeking (those with high scores tend to have high sensory thresholds and seek sensory experiences or explore their environment); Avoiding (those with high scores tend to have low sensory thresholds, retreat from sensory experiences, and require structure and predictability to tolerate them); Registration (those with high scores tend to have a high sensory threshold but respondents are passive in seeking out experiences and so can struggle to get their sensory needs met); and Sensitivity (Sensor). Individuals with high Sensitivity scores have low thresholds for sensory experiences and are passive in seeking additional sensory information from their environment (Brown & Dunn, 2002). In this way, individuals with high Sensitivity scores can react more quickly and intensely to the stimuli around them or be more aware of small changes to the environment. Sensitivity was selected for inclusion here for its closeness to sensory over-responsivity, a concept particularly linked to anxiety in research by Ben-Sasson et al. (2009) and Liss et al. (2008). Increased sensory sensitivity predicting increased anxiety is supported by previous research demonstrating that increased sensory over-responsivity (sensory sensitivity) was related to increased anxiety; for example, Black et al. (2017) found this link to be ASD specific when compared to a neurotypical population.

In Study 2, sensory sensitivity was predictive of DBC Anxiety for the teacher data, but not for the parent data. This is another example of how individuals with ASD may present differently in different contexts, particularly as pertains to their behaviour and possibly to psychiatric symptoms (Adams et al., 2018; Adams et al., 2019; Kanne et al., 2009). In terms of sensory experiences, the school environment may represent a particular challenge to individuals, especially when compared to home. Fernández-Andrés et al. (2015) found that individuals with ASD reported different sensory profiles at home compared to at school, with teachers noting higher levels of dysfunctional sensory processing. While not specifically looking at psychiatric symptoms, Fernández-Andrés et al. did note this difference was ASD specific and was not replicated by the neurotypical control group.

It is interesting to note that sensory sensitivity was not predictive of anxiety on both measures, suggesting that there is something about the nature of the measures which can impact the outcome of some relationships. The relationship between sensory sensitivity and anxiety was only found with DBC Anxiety and not with NCBRF Anxious, indicating that it may be more related to behavioural anxiety than to language- and performance-based anxiety. It may also be the case that, given the difficulties previously mentioned with measuring sensory issues with the SSP2 and AASP, this relationship may be impacted by the measurement tool used. Either way, given the close accordance of sensory issues with an ASD diagnosis, as well as the current outcome and previous research finding significant relationships between anxiety and sensory issues, this continues to be a relationship worth investigating further.

Repetitive Sensory Motor Movements. In the teacher data, increased repetitive motor movements predicted decreased anxiety on the NCBRF Anxious measure. This indicates that higher anxiety scores were associated with lower scores on the RBQ Motor, capturing movements such as pacing, rocking, twirling, and hand mannerisms. This finding was inconsistent with the suggestions of Wigham et al. (2014) and Joyce et al. (2017), that increased motor movements would be associated with heightened anxiety in individuals with ASD. It is, however, consistent with suggestions that sensory motor movements have a complex array of motivations that can lead to an inconsistent relationship with anxiety (Joosten & Bundy, 2010; Joosten et al., 2009; Russell et al., 2019). As increased levels of sensory motor activity have such a variable relationship with anxiety, repetitive sensory motor movements do not currently make a good marker for anxiety with this population. However, it may be the case that with more ASD-specific anxiety measures, a more consistent relationship would be found.

Missing Significant Variable: Self-Injury

In Study 2, six variables or observable traits were identified to be investigated as possible behavioural markers of anxiety in individuals with ASD and ID. BMA was used to create behavioural models with probability of inclusion within the regression replacing traditional *p* values to denote significance. Through this process, five of the six variables were found to be significant within at least one model; however, self-injury was consistently found to not be significant across the most probable models and regressions.

Self-injury was included within Study 2 due to its previously reported relationship with anxiety in research (Cervantes & Matson, 2015; S. Williams et al., 2015). The dynamic model of autism also included self-injury at the tertiary level for the development of anxiety in individuals with ASD (Bartak et al., 2006). It is therefore particularly interesting that it was not found to be a significant variable for inclusion in any of the behavioural models of anxiety created through the Bayesian analysis. This may have been due to differences between the measurement tools used to consider self-injury in the previous research and the current study. Cervantes and Matson (2015) used the Diagnostic Assessment for the Severely Handicapped-II, a measure created for adults with an ID. S. Williams et al. (2015) used the Behaviour Problems Inventory-short form, also created for individuals with an ID. Within the current study, the NCBRF Self-Injury subscale was used to measure self-injury, and this lack of consistency with the previous research may account for the lack of findings in the current research.

In conclusion, BMA was used to create probable models for the behavioural manifestation of anxiety in individuals with ASD and co-occurring ID. Several independent variables were identified as potential behavioural markers associated with elevated anxiety, most notably problem behaviour, insistence on sameness, and sensory sensitivity. Of note are the differences between the anxiety measures, DBC Anxiety and NCBRF Anxious, and how appropriate measure selection may be having an impact on the outcome of results and conclusions being drawn from those results. The models are considered in light of the dynamic model of autism (Bartak et al., 2006).

Theories of Anxiety: A Behavioural Model of Anxiety

The dynamic model of autism (Bartak et al., 2006) was selected for investigation in this research from a range of theoretical frameworks, including several which were ASD specific, as outlined in Chapter 4. In Study 2, behavioural models associated with anxiety in individuals with ASD and ID were created. These behavioural models will now be considered in relation not only to the dynamic model of autism, but also to the other ASD-specific anxiety theories, namely the theories offered by Wood and Gadow (2010), Gotham et al. (2013), and Kerns et al. (2014), to see if they can explain the manifestation of anxiety in individuals with ASD and ID.

Study 2 was an exploratory study that used prior research, including the findings from Study 1, to identify the most relevant independent variables, and then created the highest probability models for predicting elevated anxiety. Four models were produced, with the parent data models being simplified versions of the teacher data models, with fewer significant independent variables and no relationships additional to those within the teacher data. The behavioural model created in Study 2 with NCBRF Anxious teacher data found four variables to be predictive of increased anxiety: increased problem behaviour, increased insistence on sameness, increased social communication, and decreased motor repetitive behaviours. The model created with DBC Anxiety teacher data also found increased problem behaviour and insistence on sameness to be significant variables, as well as increased sensory sensitivity. The teacher models for each of the anxiety measures will be considered in more detail here in relation to existing models of anxiety.

The behavioural models were only partially supported by Wood and Gadow's (2010) mood dysregulation and anxiety theory, which focuses on social confusion, peer rejection, prevention of repetitive behaviours, and aversive sensory experiences, leading to the experience of mood dysregulation and anxiety and resulting in an array of effects for the individual. Wood and Gadow's anxiety model specifies outcomes such as increased social avoidance, increased ASD symptoms (e.g., RRBs), problem behaviours, and personal distress. Motor behaviour (decreased levels of which was predicted under NCBRF Anxious) cannot be included under increased ASD symptoms, but problem behaviour, social communication, and insistence on sameness could be captured under this theory. In terms of DBC Anxiety, both increased problem behaviour and insistence on sameness can be included, but increased sensory sensitivity cannot, as there is no specific provision for sensory sensitivity within the Wood and Gadow model.

In addition to these two variables not having a place within Wood and Gadow's (2010) theory, there is also the larger issue of relevance for those with co-occurring ID. As was identified in Chapter 4, the theory's focus on social functioning and cognitive processing of experiences as the only means of developing anxiety makes it inapplicable to those with ASD and ID, despite some of the relevant behavioural indicators being present. As a result, Wood and Gadow's anxiety theory does not offer a reasonable explanation for the development of anxiety in individuals with ASD and ID.

Gotham et al.'s (2013) theory investigated insistence on sameness specifically, differentiating it from anxiety and from other repetitive behaviours; it does not directly relate to the expression of anxiety. Within this model, it was therefore not a behavioural marker for anxiety; nor were increased ASD symptoms such as increased social communication difficulties or decreased motor behaviours (both of which are predicted by NCBRF Anxious). Sensory sensitivity is also not included within the model. As such, while many similar variables exist within the Gotham et al. model, the only behaviour marker they directly relate to the expression of anxiety is nonspecific problem behaviour. This model therefore offers limited insight into the behavioural models of anxiety found in Study 2.

Another key issue with this theory, however, is the finding as part of the Gotham et al. (2013) research that increased anxiety was associated with no ID. As such, Gotham et al.'s anxiety theory does not offer a reasonable explanation for how anxiety develops in an individual with ASD and co-occurring ID.

The behavioural models are difficult to interpret within Kerns et al. (2014) theory of anxiety for individuals with ASD, which considers both traditional and atypical anxiety presentation. They suggested that traditional anxiety can exist and follow typical DSM criteria and diagnostic patterns, being quite separate from ASD symptoms and most likely being associated with higher IQ and language ability. By contrast, atypical anxiety may be more individually driven, overlapping more with ASD symptomology and possibly with higher anxiety levels. As such, the significant variables from the behavioural models created in Study 2 that are associated with increased ASD symptomology, such as reduced social communication and insistence on sameness, can be explained within Kerns et al.'s atypical anxiety theory. Hypersensitivity is listed as a symptom of traditional anxiety, and could be seen as similar to increased sensory sensitivity from the DBC Anxiety behaviour model. However, problem behaviour is not identified as characteristic of either traditional or atypical anxiety within this theory, and the finding of decreased motor repetitive behaviours does not fit into Kerns et al.'s theory. It is also of note that the anxiety measure used by Kerns et al. was developed for a neurotypical population and the research itself included only individuals with a mild ID, so those with a more significant ID were not considered. As such, this theory is of little relevance or utility for those with ASD and a more significantly impacting ID.

The dynamic model of autism (Bartak et al., 2006) was selected for investigation as it offered an explanation for the development of anxiety in individuals with ASD and a co-occurring ID. This theory was supported in Study 1 through a series of multiple regressions. In relation to Study 2, the dynamic model of autism postulates that the primary deficits of ASD are social communication and information processing difficulties such as executive functioning, central coherence, and theory of mind; these interact with environmental stressors to create secondary impacts, which are likely to be coping strategies. Secondary impacts include increased ritualistic/repetitive behaviours (such as insistence on sameness), poor sensory integration, and odd social behaviours. Stress is continually experienced by the individual; intervention often focuses on reducing the secondary features of ASD, which in turn leads to challenging or problem behaviour, self-injury, or internalising behaviours. Continued stress and lack of support can lead to psychiatric conditions, including anxiety disorders.

The NCBRF Anxious teacher data behavioural model found that decreased social communication difficulties (decreased ASD symptoms) at the primary level led to decreased sensory motor movements and increased insistence on sameness at the secondary, then increased problem behaviour at the tertiary level, which in turn led to heighted anxiety. This pattern is not consistent with the overall pattern predicted by the dynamic model of autism (Bartak et al., 2006). As the theoretical framework has been validated in Study 1, and different results have been found with another anxiety measure, this finding puts further emphasis on ensuring the anxiety measurement tool is appropriate for the population in question.

The behavioural model produced using DBC Anxiety teacher data found three significant independent variables, which likewise can be assigned to the levels of the dynamic model of autism (Bartak et al., 2006). This model, however, lacks an entry at the primary level, and so assumes all participants are similarly diagnosed in terms of their ASD and ID; that is, that they all experience significant levels of difficulties with their receptive and expressive communication, considerable deficits in theory of mind and executive functioning, and weak central coherence. When encountering environmental stressors, secondary level behaviours emerge from within this behavioural model: increased insistence on sameness and increased sensory sensitivity. As stressors continue to be experienced, increased problem behaviours emerge at the tertiary level. A lack of support and understanding while experiencing continued stress leads to heightened anxiety at the quaternary level. This behavioural model is consistent with the dynamic model of autism and offers some real insight into how anxiety develops in individuals with ASD and ID, as well as in to how it can be identified though observable behaviours.

The aim of Studies 1 and 2 was to investigate a theoretical framework that explains how anxiety develops in individuals with ASD and co-occurring ID, and then to determine if behavioural patterns exist that could help identify anxiety within those same individuals. In Study 1, the dynamic model of autism (Bartak et al., 2006) was found to provide insight into how anxiety develops in individuals with ASD and ID, with problem behaviour found to be a significant independent variable of interest for further investigation. In Study 2, behavioural manifestation models of anxiety were created using the BMA method, identifying a number of variables associated with elevated anxiety. In considering the behavioural models through the lens of each of the ASD-specific anxiety theories available, the dynamic model of autism offers the most relevant explanation to understand how anxiety develops in individuals with ASD and ID.

Clinical Implications

The dynamic model of autism (Bartak et al., 2006) offers a framework to reconceptualise how anxiety, ASD, and ID intersect. By better understanding how these complex phenomena interact, the dynamic model of autism allows us to interpret behavioural escalations as symptoms of anxiety, and then use the model itself as a tool in considering behavioural de-escalation and the treatment of anxiety. This could result in better resolved behavioural escalations with longer lasting outcomes for individuals with ASD and co-occurring ID.

Current behaviour support practice has been to target problem or challenging behaviour, generally with functional assessments, which do not consider the behaviour as symptoms or as part of a more complex mental health condition. Instead, the behaviour is thought to be maladaptive and needing to be extinguished or replaced, with thought given to a communicative message, but not necessarily to an underlying condition or cause. Treatment of anxiety in a more targeted way may therefore result in longer term stability. This is particularly important given the prevalence of challenging and problem behaviour in this population and the current lack of success with long-term treatment. Murphy et al. (2009) reported that 82% of people with ASD and ID displayed at least one challenging behaviour; 96% more than one. Davies and Oliver (2014), Oliver et al. (2017), and Podlesnik and Kelley (2017) all reported that challenging behaviour (and self-injury in particular) are persistent behaviours that often relapse or reinstate despite intensive behavioural intervention.

The dynamic model of autism (Bartak et al., 2006) also offers an opportunity for treatment or intervention options. An individual with ASD and ID and a significant anxiety disorder is likely to also have increased problem behaviour, sensory sensitivities, and repetitive behaviours (insistence on sameness). This is likely to present as general agitation and overarousal, making the individual hard to manage and also inaccessible for many skills- or strategies-based learning programs designed to aid in recovery from anxiety. Given the complex behavioural picture presented by an individual with ASD and ID, there is a need to consider multimodal elements from multiple fields to best support that individual. Treatment of anxiety could then best be approached by stepping back through the levels of the dynamic model of autism. This may, in turn, make individuals with ASD, ID, and an anxiety disorder more available to learning skills and alternative coping strategies, generally, by first addressing anxiety and de-escalating arousal levels. Then, as their skills develop, even slowly over time, the likelihood of relapse is reduced, and skills development is increased. For example, if an individual can increase their communication skills to express a wider range of needs, using an augmented communication device such as an iPad or through visuals, then their frustration in the future may also be reduced.

An example of a multimodal approach to supporting anxiety has been applied by Moskowitz, Walsh, et al. (2017) when treating anxiety in their research. They used individualised elements of cognitive behaviour therapy (e.g., graduated exposure, psychoeducation, cognitive restructuring), positive behaviour support (e.g., visual schedules, providing choices, more concrete language use, incorporating special interests), and applied behaviour analysis (e.g., contingent reinforcement, differential reinforcement, escape extinction) in their study of individuals with ASD and ID. There were limitations in their research, however, due to their focus on specific situations of heightened anxiety (such as phobias, that can be addressed through exposure or a planned schedule) rather than more generalised anxiety states.

Currently, there are limited anxiety treatment options for individuals with ASD and co-occurring ID. Several ASD-specific cognitive behaviour therapy programs have been developed over the last 10 years, with some success reported (Hepburn et al., 2016; McConachie et al., 2014; Rodgers, Hodgson, et al., 2016). A single-group treatment program was found which targeted anxiety for individuals with ASD and mild ID (Blakeley-Smith et al., 2021), in addition to several case studies focusing on individuals with ASD and various levels of ID (Kildahl et al., 2021; Moskowitz, Rosen, et al., 2017). However, even if anxiety is correctly identified in the first place for individuals with ASD and more substantially impacting ID, there are few treatment options for their anxiety.

Behavioural escalations in individuals with ASD and ID are commonly identified by parents and teachers and can persist long-term and cause considerable harm and risk to individuals. Addressing problem behaviour is difficult: research has shown that gains from direct interventions can be followed by relapse or reinstatement of behaviours. By considering the possible underlying cause of the behaviours as anxiety and treating problem behaviour as a symptom rather than as the target, intervention and treatment programs may become more successful.

Limitations of the Current Research

Study 1 and Study 2 were exploratory in nature, testing the dynamic model of autism (Bartak et al., 2006) to see if it provides an explanation for the development of anxiety in individuals with ASD and ID and potentially informs behavioural models predicting elevated anxiety in those same individuals. The participant numbers in this research and the resulting power of the statistics limit the generalisability of the results to the broader ASD and ID population. As a result, replication of both Study 1 and 2 in a larger group is recommended.

It is perhaps this limited participant pool that resulted in a lack of normal distribution for several variables. This was a particular issue in Study 1, and several measures were taken to address skewness and kurtosis and to account for residual distributions, linearity, and homoscedasticity. Although the ASD and ID population is heterogenous by nature, normal distribution may have occurred with a larger participant pool. The population size nonetheless limits our ability to generalise the findings beyond this initial exploratory study.

Anxiety Measurement Tools

As discussed consistently in this thesis, choice of anxiety measurement tool has been found to have a significant impact on the outcome of the data analysis and the resulting conclusions. This has been found to be a particular issue for the under-researched ASD and co-occurring ID population, for whom no specific anxiety measure has been developed.

The DBC Anxiety and NCBRF Anxious were chosen as dependent variables in Studies 1 and 2 as they utilise different markers of anxiety. The DBC Anxiety used behaviourally based items, while the NCBRF Anxious used more language- and performance-based anxiety items. There was no overlap in terms of individual items between the two measures. In this study, the DBC Anxiety and NCBRF Anxious were found to be only moderately correlated with each other for both the parent and teacher data. Research comparing the NCBRF and DBC directly is limited. Norris and Lecavalier (2011) completed an evaluation of the validity of the NCBRF using the DBC as a comparison, reporting significant correlations in the expected directions. However, the correlation between the DBC Anxiety and the NCBRF Anxious subscales was particularly small (r = .39), when parent-reported data was used for children aged 5 to 18 with a variety of developmental disability diagnoses. These two anxiety measures are therefore not interchangeable, and it is doubtful they are testing the same phenomena given the lack of correlation between them. If the dependent variable is not measuring anxiety for the test population, this will have a considerable impact on the outcome. This is particularly highlighted by the different outcomes found in the multiple regressions in Study 1 when different anxiety measures were used as the dependent variable. The results for the two scales are inconsistent, suggesting that not all measurement tools are well suited to the population being investigated.

The lack of well-validated anxiety measures for those with a diagnosis of ASD is noted in the research (Grondhuis & Aman, 2012; MacNeil et al., 2009; Matson & Cervantes, 2014; Moskowitz, Rosen, et al., 2017). Due to the difficulties diagnosing anxiety in those with ASD, and the reliance on gathering information from others in this process, there has been a growing recognition of the need to develop ASD-specific anxiety measures (Appleton et al., 2019; Toscano et al., 2020). As such, several have been created in the last few years, notably the Anxiety Scale for Children with Autism Spectrum Disorder (ASC-ASD) by Rodgers, Wigham, et al. (2016), the Anxiety Disorders Interview Schedule – Autism Spectrum Addendum (ADIS/ASA) by Kerns et al. (2014), and the Macquarie Anxiety Behavioural Scale (MABS) by Toscano et al. (2020). There has also been a recent a move to validate the more widely used Depression, Anxiety and Stress Scales (DASS) by Park et al. (2020) for use with individuals with ASD; however, the measure is specifically for ASD individuals without an ID. These ASD-specific anxiety measures all include items that require a high level of cognition or recognition of internal experience, language ability, and social communication. While some do include observable traits and behavioural elements, there is still a lack of consideration of the broader ASD spectrum, particularly those with a co-occurring ID.

Mingins et al. (2021) reviewed existing research into individuals with ASD, ID, and anxiety, and found that the lack of validated anxiety measures for this population was the most notable research limitation. Mingins et al. also noted that this lack of consistency limits the conclusions that can be drawn from research, as few comparisons can be drawn between data sets when such different anxiety measurement tools are used.

Given this need for ASD-specific measures, it is of note that to date the DBC and NCBRF used in this research have not been normed for an ASD and ID population. The DBC has been used widely in ASD and ID research and has normative data covering a range of ID levels. The DBC also contains an early screener to identify risk of an ASD diagnosis (Einfeld & Tonge, 1995). However, the DBC Anxiety subscale contains only eight questions that have little crossover with any of the items on the NCBRF Anxious, as outlined in Chapter 5. Appleton et al. (2019) and Mingins et al.

(2021) have pointed out the lack of consistency between existing anxiety measures, which limits our ability to draw comparisons and conclusions, even if the populations were more homogenous. The limitation of measurement tools currently available to capture anxiety within this population is therefore significant.

Future Directions

The dynamic model of autism (Bartak et al., 2006) was investigated in Study 1. The model was found to offer some explanation for the development of anxiety in individuals with ASD and ID. The theoretical framework was clinically driven and had not previously been empirically tested. Given the high incidence of anxiety within the ASD population and the possible insights the dynamic model of autism offers in terms of understanding how core characteristics of ASD create a vulnerability for developing anxiety, it is worth further investigation. Given this is an exploratory study, further validation of the dynamic model of autism through additional research with individuals with ASD and ID should be considered. Of key concern is the anxiety measure used as a dependent variable in future research. As discussed, this can have a considerable impact on the outcomes found and conclusions reached. As the dynamic model of autism potentially has utility across the autism spectrum, it would also be interesting to investigate the model in relation to the full ASD spectrum of individuals with and without ID and with a wider range of social communication and language abilities. This would allow the model itself to be more broadly tested for applicability to the full spectrum of those diagnosed with ASD. Special consideration should be given to the utilisation of ASD-specific anxiety measures, which may be more appropriately used to collect information for this population.

Individuals with ASD and co-occurring ID are, to date, an under-researched and possibly underdiagnosed population. Behavioural patterns have the potential to provide a more observable set of markers for identifying anxiety in this population. The current research was exploratory, but consideration of broader behavioural patterns as markers for a psychiatric syndrome such as anxiety appears to have merit and should be further investigated to confirm the current research and to see if additional observable traits could be incorporated into a behavioural model for anxiety for individuals with ASD and ID.

Anxiety was measured within this study, at a single point of time, capturing how the participants presented at one point of reference. However, as was discussed in Chapter 4, stress and anxiety may be better conceptualised as a continuum rather than as a binary variable. An individual's experience of stress and anxiety may shift continually due to numerous factors. This would certainly be true considering the fluctuating nature of anxiety and the multiple variables involved with the dynamic model of autism (Bartak et al., 2006). Most anxiety research has focused on anxiety at a single point of reference (K. Dunn et al., 2020; Helverschou & Martinsen, 2011; Hess, 2009; Joosten et al., 2009; Kerns et al., 2017; Lecavalier, 2006; Sukhodolsky et al., 2008). Moskowitz et al. (2013), by contrast, identified high-stress situations and completed their anxiety measurements within those situations (multimodal, including physiological response to anxiety, problem-behaviour-specific and anxiety-specific behaviours). Several researchers have noted the individual differences present in those with ASD and anxiety and the need to consider multiple factors in the diagnostic process (MacNeil et al., 2009; Moskowitz, Rosen, et al., 2017). Given these issues and possible fluctuations due to numerous variables, anxiety may be more accurately considered as changing over time from a baseline, rather than as a binary entity. A longitudinal case design may offer valuable insight into how anxiety develops and changes in individuals with ASD and ID, assisting with the identification of early warning signs such as increases in insistence on sameness or sensory sensitivity. These indicators may enable an earlier entry into treatment. Early treatment, before entrenched problem behaviours or mental ill health have had an opportunity to develop, is likely to be more successful and to require less resources.

As discussed, the lack of a relationship between self-injury and anxiety within this research is of particular note. This may have been due to the measurement tool used, the NCBRF SIB/Stereotypic, which is a Likert scale 0–3 with seven items. This offers a very limited scale both in terms of item number and scale of intensity. Also, in this form of data collection for self-injury, the true impact of chronic and dangerous behaviour may be omitted. Questionnaires for self-injury are generally used to rate and then tally the frequency of three or four behaviours. Clinical experience would suggest that individuals with ASD and ID with significant levels of self-injury (that is, they engage in the behaviour numerous times per day, or with high intensity less frequently), are likely to have a dominant self-injurious behaviour, such as self-biting. At times of very high distress this may change to include other behaviours such as hair pulling or head hitting, but typically there is a single frequent behaviour that is accessed regularly.

The primary behaviour of self-injury can also change with time, as described by Kildahl et al. (2021) in their case study of an individual with ASD, ID, and anxiety: As a young child the individual displayed hand-biting behaviours, but this changed to hitting their head against walls and the floor as they got older. No other research was found to support this observation of self-injury specificity or its changing nature over time. Such variability reduces the capacity of a questionnaire format capturing meaningful information about self-injurious behaviour for research purposes, as even a Likert scale of 0–3 will have limited impact for a single behaviour. As Davies and Oliver (2016) and Oliver et al. (2017) reported, self-injurious behaviours tend to persist for several years. A measurement tool that is sensitive to the frequency, persistence, and damage caused by the behaviour would allow for more accurate information to be gathered about the true impact of self-injurious behaviour in individuals with ASD and

ID. Podlesnik and Kelley (2017) reported that self-injurious behaviour in individuals with ASD was highly likely to recur even after intensive intervention had extinguished the behaviour. Perhaps if we better understand the relationship between SIB and anxiety and the driving forces behind SIB, the behaviour could more effectively be addressed and reduced and the safety and quality of life for many individuals with ASD and ID increased.

Conclusion

In this thesis the complex relationship that exists between anxiety, ASD, and ID was considered. The published scoping review (Appleton et al., 2019) identified the paucity of research in this area, particularly when considering how we identify and diagnose anxiety within this unique population. A range of issues with the diagnostic process were identified through both the scoping review and further literature review in Chapters 2 and 3, with the core features and information processing patterns of ASD and cognitive delays inherent in ID both playing substantial roles in leading to underdiagnosis of anxiety, and a lack of treatment options.

Of the anxiety theories reviewed (Chapter 4), only the dynamic model of autism (Bartak et al., 2006) was found to be relevant to understanding how anxiety develops in those with ASD and ID. This theory was the focus of Study 1. With data from a group of children and young adults with ASD and ID, the dynamic model of autism was found to offer a theoretical framework for the development of anxiety in this population, with problem behaviour being a consistent predictive variable regardless of the anxiety measure used or the data source. Given the significant issues with traditional diagnostic techniques for this population, there is a need to focus on broader behavioural patterns associated with anxiety to aid with identification. Study 2 created behavioural models that were most predictive of anxiety, using a combination of previous research as well as the outcomes of Study 1 to inform the selection of variables. As a result, increased problem behaviour, increased insistence on sameness, and increased sensory sensitivity were variables most predictive of increased anxiety. The consideration that anxiety may be an underlying cause of increased problem behaviour and ASD symptomology (such as RRBs like insistence on sameness) offers more meaningful ways of treating and addressing these otherwise poorly supported issues for individuals with ASD and ID.

A key finding from this research was the lack of population-specific measurement tools, particularly in relation to measuring anxiety. No anxiety questionnaire could be found that was created or validated for individuals with ASD and ID, despite research indicating the uniqueness of this population. Both Studies 1 and 2 found divergent results based on the different anxiety measurement tools used, potentially significantly altering the conclusions drawn. This highlights the need for appropriate measurement tools to be chosen for the population being studied.

The issue of how anxiety manifests and is identified in individuals with ASD and ID is complex and multifaceted. This exploratory research offers insight in to the need for further research to broaden our understanding of this intricate subject and the way this under-researched population requires more adequate support. If prevalence rates are established and acknowledged in research and a better understanding of behavioural manifestations developed, effective treatment pathways can be established.

References

Adams, D., Clark, M., & Keen, D. (2019). Using self-report to explore the relationship between anxiety and quality of life in children on the autism spectrum. *Autism Research, 12*(10), 1505–1515. https://doi.org/10.1002/aur.2155

Adams, D., Simpson, K., & Keen, D. (2018). School-related anxiety symptomatology in a community sample of primary-school-aged children on the autism spectrum. *Journal of School Psychology, 70*, 64–73. https://doi.org/10.1016/j.jsp.2018.07.003

Aman, M., Tasse, M. J., Rojahn, J., & Hammer, D. (1996). The Nisonger CBRF: A child behavior rating form for children with developmental disabilities. *Research in Developmental Disabilities, 17*(1), 41–57. https://doi.org/10.1016/0891-4222(95)00039-9

American Psychiatric Association. (1987). *Diagnostic and statistical manual of mental disorders* (3rd TR ed.).

American Psychiatric Association. (1994). *Diagnostic and statistical manual of mental disorders* (4th ed.).

American Psychiatric Association. (2000). *Diagnostic and statistical manual of mental disorders* (4th TR ed.).

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). American Psychiatric Publishing.

Appleton, H., Roberts, J., & Simpson, K. (2019). How is anxiety identified and diagnosed in individuals with autism spectrum disorder and intellectual disability? A scoping review. *Journal of Mental Health Research in Intellectual Disabilities, 12*(3–4), 152–175. https://doi.org/10.1080/19315864.2019.1679299

Bailey, N. M., & Andrews, T. M. (2003). Diagnostic criteria for psychiatric disorders for use with adults with learning disability/mental retardation (DC-LD) and the diagnosis of anixety disorders: A review. *Journal of Intellectual Disability Research, 47*, 50–61. https://doi.org/10.1046/j.1365-2788.47.s1.25.x

Baio, J., Wiggins, L., Christensen, D. L., Maenner, M. J., Daniels, J., Warren, Z., Kurzius-Spencer, M., Zahorodny, W., Rosenberg, C. R., White, T., Durkin, M. S., Imm, P., Nikolaou, L., Yeargin-Allsopp, M., Lee, L.-C., Harrington, R., Lopez, M., Fitzgerald, R. T., Hewitt, A. . . . Dowling, N. F. (2018). Prevalence of autism spectrum disorder among children aged 8 years - autism and developmental disability monitoring network, 11 sites United States, 2014. Morbidity and mortality weekly report. *Surveillance Summaries, 67*, 1–23. https://www.ncbi.nlm.nih.gov/pmc/articles/PMC5919599/pdf/ss6706a1.pdf

Bar-Haim, Y., Lamy, D., Pergamin, L., Bakermans-Kranenburg, M. J., & van Ijzendoorn, M. H. (2007). Threat-Related attentional bias in anxious and nonanxious individuals: A meta-analytic study. *Psychological Bulletin, 133*(1), 1–24. https://doi.org/10.1037/0033-2909.133.1.1

Baron-Cohen, S., Leslie, A. M., & Frith, U. (1985). Does the autistic child have a “theory of mind”? *Cognition, 21*(1), 37–46. https://doi.org/10.1016/0010-0277(85)90022-8

Bartak, L., Bottroff, V., & Zeitz, J. (2006). Therapist insights in working with stress in people with autism spectrum disorder. In M. G. Baron, J. Groden, G. Groden, & L. Lipsitt (Eds.), *Stress and coping in autism* (pp. 246–273). Oxford University Press.

Bauer, A. M., Quas, J. A., & Boyce, W. T. (2002). Associations between physiological reactivity and children's beahviour: Advantages of a multisystem approach. *Developmental and Behavioral Pediatrics, 23*(2), 102–113. https://doi.org/0196-206X/00/2302-0102

Baxter, A. J., Brugha, T. S., Erskine, H. E., Scheurer, R. W., Vos, T., & Scott, J. G. (2015). The epidemiology and global burden of autism spectrum disorders. Psychological Medicine, 45(3), 601–13. https://doi.org/10.1017/S003329171400172X

Beck, A. T. (1979). *Cognitive therapy and the emotional disorders*. New American Library.

Ben-Sasson, A., Carter, A. S., & Briggs-Gowan, M. J. (2009). Sensory over-responsivity in elementary school: Prevalence and social-emotional correlates. *Journal of Abnormal Child Psychology, 37*, 705–716. https://doi.org/10.1007/s10802-008-9295-8

Ben-Sasson, A., Cermak, S. A., Orsmond, G. I., Tager-Flusberg, H., Kadlec, M. B., & Carter, A. S. (2008). Sensory clusters of toddlers with autism spectrum disorders: Differences in affective symptoms. *The Journal of Child Psychology and Psychiatry, 49*(8), 817–825. https://doi.org/10.1111/j.1469-7610.2008.01899.x

Berument, S. K., Rutter, M., Lord, C., Pickles, A., & Bailey, A. (1999). Autism screening questionnaire: Diagnostic validity. *The British Journal of Psychiatry, 175*(5), 444–451.

Bishop-Fitzpatrick, L., Minshew, N. J., & Eack, S. M. (2013). A systematic review of psychosocial interventions for adults with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 43*(3), 687–694. https://doi.org/10.1007/s10803-012-1615-8

Bitsika, V., & Sharpley, C. F. (2016). Which aspects of challenging behaviour are associated with anxiety across two age groups of young males with an autism spectrum disorder? *Journal of Developmental and Physical Disabilities, 28*(5), 685–701. https://doi.org/10.1007/s10882-016-9502-4

Bitsika, V., Sharpley, C. F., Sweeney, J. A., & McFarlane, J. R. (2014). HPA and SAM axis responses as correlates of self-vs parental ratings of anxiety in boys with an autistic disorder. *Physiology & Behavior, 127*, 1–7. https://doi.org/10.1016/j.physbeh.2013.12.011

Black, K. R., Stevenson, R. A., Segers, M., Ncube, B. L., Sun, S. Z., Philipp-Muller, A., Bebko, J. M., Barense, M. D., & Ferber, S. (2017). Linking anxiety and insistence on sameness in autistic children: The role of sensory hypersensitivity. *Journal of Autism and Developmental Disorders, 47*(8), 2459–2470. https://doi.org/10.1007/s10803-017-3161-x

Blakeley-Smith, A., Meyer, A. T., Boles, R. E., & Reaven, J. (2021). Group cognitive behavioural treatment for anxiety in autistic adolescents with intellectual disability: A pilot and feasibility study. *Journal of Applied Research in Intellectual Disabilities, 34*(3), 777–788. https://doi.org/10.1111/jar.12854

Bradley, E. A., Summers, J. A., Wood, H. L., & Bryson, S. E. (2004). Comparing rates of psychiatric and behavior disorders in adolescents and young adults with severe intellectual disability with and without autism. *Journal of Autism and Developmental Disorders, 34*(2), 151–161. https://doi.org/10.1023/B:JADD.0000022606.97580.19

Brady, E. U., & Kendall, P. C. (1992). Comorbidity of anxiety and depression in children and adolescents. *Psychological Bulletin, 111*, 244–255.

Brereton, A. V., Tonge, B. J., & Einfeld, S. L. (2006). Psychopathology in children and adolescents with autism compared to young people with intellectual disability. *Journal of Autism and Developmental Disorders, 36*(7), 863–870. https://doi.org/10.1007/s/10803-006-0125-y

Brooks, W. T., & Benson, B. A. (2013). The validity of the Social Communication Questionnaire in adults with intellectual disability. *Research in Autism Spectrum Disorders, 7*(2), 247–255. https://doi.org/10.1016/j.rasd.2012.10.002

Brown, C., & Dunn, W. (2002). *Adolescent/Adult Sensory Profile User's Manual*. Pearson.

Burns, C. O., Dixon, D. R., Novack, M., & Granpeesheh, D. (2017). A systematic review of assessments for sensory processing abnormalities in autism spectrum disorder. *Review Journal of Autism and Developmental Disorders, 4*(3), 209–224. https://doi.org/10.1007/s40489-017-0109-1

Cannon, M. F., & Weems, C. F. (2006). Do anxiety and depression cluster into distinct groups?: A test of tripartite model predictions in a community sample of youth. *Depression and Anxiety, 23*(8), 453–460. https://doi.org/10.1002/da.20215

Cashin, A., & Yorke, J. (2018). The relationship between anxiety, external structure, behavioral history and becoming locked into restricted and repetitive behaviors in autism spectrum disorder. *Issues in Mental Health Nursing, 39*(6), 533–537. https://doi.org/10.1080/01612840.2017.1418035

Cervantes, P. E., & Matson, J. L. (2015). Comorbid symptomology in adults with autism spectrum disorder and intellectual disability. *Journal of Autism and Developmental Disorders, 45*(12), 3961–3970. https://doi.org/10.1007/s10803-015-2553-z

Chakrabarti, S., & Fombonne, E. (2005). Pervasive developmental disorders in preschool children: Confirmation of high prevalence. *The American Journal of Psychiatry, 162*(6), 1133–1141. https://doi.org/10.1176/appi.ajp.162.6.1133

Chang, Y.-C., Quan, J., & Wood, J. J. (2012). Effects of anxiety disorder severity on social functioning in children with autism spectrum disorders. *Journal of*

Developmental and Physical Disabilities, 24(3), 235–245. https://doi.org/10.1007/s10882-012-9268-2

Clark, L. A., & Watson, D. (1991). Tripartite model of anxiety and depression: Psychometric evidence and taxonomic implications. *Journal of Abnormal Psychology, 100*(3), 316–336.

Cole, D. A., Truglio, R., & Peeke, L. (1997). Relation between symptoms of anxiety and depression in children: A multitrait-multimethod-multigroup assessment. *Journal of Consulting and Clinical Psychology, 65*(1), 110–119. https://doi.org/10.1037/0022-006X.65.1.110

Craske, M. G., Rauch, S. L., Ursajo, R., Prenoveau, J., Pine, D. S., & Zinbarg, R. E. (2009). What is an anxiety disorder? *Depression and Anxiety, 26*, 1066–1085. https://doi.org/10.1002/da.20633

Cuccaro, M. L., Shao, Y., Grubber, J., Slifer, M., Wolpert, C. M., Donnelly, S. L., Abramson, R. K., Ravan, S. A., Wright, H. H., DeLong, G. R., & Pericak-Vance, M. A. (2003). Factor analysis restricted and repetitive behaviours in autism using the Autism Diagnostic Interview-R. *Child Psychiatry and Human Development, 34*(1), 3–17. https://doi.org/10.1023/A:1025321707947

Davies, L. E., & Oliver, C. (2016). Self-injury, aggression and destruction in children with severe intellectual disability: Incidence, persistence and novel, predictive behavioural risk markers. *Research in Developmental Disabilities, 49-50*, 291–301. https://doi.org/10.1016/j.ridd.2015.12.003

Davis, T. E., Fodstad, J. C., Jenkins, W. S., Hess, J. A., Moree, B. N., Dempsey, T., & Matson, J. L. (2010). Anxiety and avoidance in infants and toddlers with autism spectrum disorders: Evidence for differing symptom severity and presentation. *Research in Autism Spectrum Disorders, 4*(2), 305–313. https://doi.org/10.1016/j.rasd.2009.10.002

Davis, T. E., Moree, B. N., Dempsey, T., Hess, J. A., Jenkins, W. S., Fodstad, J. C., & Matson, J. L. (2012). The effect of communication deficits on anxiety symptoms in infants and toddlers with autism spectrum disorders. *Behavior Therapy, 43*(1), 142–152. https://doi.org/10.1016/j.beth.2011.05.003

Davis, T. E., Moree, B. N., Dempsey, T., Reuther, E. T., Fodstad, J. C., Hess, J. A., Jenkins, W. S., & Matson, J. L. (2011). The relationship between autism spectrum disorders and anxiety: The moderating effect of communication. *Research in Autism Spectrum Disorders, 5*(1), 324–329. https://doi.org/10.1016/j.rasd.2010.04.015

Deb, S., & Prasad, K. (1994). The prevalence of autistic disorder among children with a learning disability. *British Journal of Psychiatry, 165*, 395–399. https://doi.org/10.1192/bjp.165.3.395

de Marchena, A. B., Eigsti, I.-M., & Yerys, B. E. (2015). Brief report: Generalization weaknesses in verbally fluent children and adolescents with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 45*(10), 3370–3376. https://doi.org/10.1007/s10803-015-2478-6

Dickerson Mayes, S., Calhoun, S. L., Murray, M. J., & Zahid, J. (2011). Variables associated with anxiety and depression in children with autism. *Journal of Developmental and Physical Disabilities, 23*, 325–337. https://doi.org/10.1007/s10882-011-9231-7

Dugas, M. J., Gagnon, F., Ladouceur, R., & Fresston, M. (1998). Generalized anxiety disorder: A preliminary test of a conceptual model. *Behaviour Research and Therapy, 36*(2), 215–226. https://doi.org/10.1016/S0005-7967(97)00070-3

Dugas, M. J., Sexton, K. A., Hebert, E. A., Bouchard, S., Gouin, J.-P., & Shafran, R. (2022). Behavioral experiments for intolerance of uncertainty: A randomized clinical trial for adults with generalized anxiety disorder. *Behavior Therapy*. https://doi.org/10.1016/j.beth.2022.05.003

Dunn, K., Rydzewska, E., Fleming, M., & Cooper, S.-A. (2020). Prevalence of mental health conditions, sensory impairments and physical disability in people with co-occurring intellectual disabilities and autism compared with other people: A cross-sectional total population study in Scotland. *BMJ Open, 10*(4), e035280-e035280. https://doi.org/10.1136/bmjopen-2019-035280

Dunn, W. (2014). *Sensory Profile 2 user's manual*. Pearson.

Dykens, E., & Lambert, W. (2013). Trajectories of diurnal cortisol in mothers of children with autism and orher develomental disabilities: Relations to health and mental health. *Journal of Autism and Developmental Disorders, 43*, 2426–2434. https://doi.org/10.1007/s10803-013-1791-1

Eeles, A., Spittle, A., Anderson, P., Brown, N., Lee, K., Boyd, R., & Doyle, L. (2013). Assessments of sensory processing in infants: A systematic review. *Developmental Medicine and Child Neurology, 55*(4), 314–326. https://doi.org/10.1111/j.1469-8749.2012.04434.x

Einfeld, S. L., & Tonge, B. J. (1995). The Developmental Behaviour Checklist: the development and validation of an instrument to assess behavioural and emotional disturbance in children and adolescents with mental retardation. *Journal of Autism and Developmental Disabilities, 25*(2), 81–104. https://doi.org/10.1007/BF02178498

Einfeld, S., & Tonge, B. J. (2002). *Developmental Behaviour Checklist (DBC).* University of New South Wales and Monash University.

Esbensen, A. J., Mailick Seltzer, M., Lam, K. S. L., & Bodfish, J. W. (2009). Age-related differences in restricted repetitive behaviours in autism spectrum disorders. *Journal of Autism and Developmental Disorders, 39*, 57–66. https://doi.org/10.1007/s10803-008-0599-x

Eysenck, M. W., & Byrne, A. (1992). Anxiety and susceptibility to distraction. *Personality and Individual Differences, 13*(7), 793–798.

Fernández-Andrés, M. I., Pastor-Cerezuela, G., Sanz-Cervera, P., & Tárraga-Mínguez, R. (2015). A comparative study of sensory processing in children with and without autism spectrum disorder in the home and classroom environments. *Research in Developmental Disabilities, 38*, 202–212. https://doi.org/10.1016/j.ridd.2014.12.034

Fombonne, E. (2018). Editorial: The rising prevalence of autism. *Journal of Child Psychology and Psychiatry, 59*(7), 717–720. https://doi.org/10.1111/jcpp.12941

Friedman, B. H., & Thayer, J. F. (1998). Anxiety and autonomic flexibility: A cardiovascular approach. *Biological Psychology, 47*(3), 243–263. https://doi.org/10.1016/S0301-0511(97)00027-6

Fries, E., Hesse, J., Hellhammer, J., & Hellhammer, D. H. (2005). A new view on hypocortisolism. *Psychoneuroendocrinology, 30*(10), 1010–1016. https://doi.org/10.1016/j.psyneuen.2005.04.006

Frith, U., & Happé, F. (1994). Autism: Beyond theory of mind. *Cognition, 50*(1–3), 115–132. https://doi.org/10.1016/0010-0277(94)90024-8

Gadow, K. D., DeVincent, C. J., & Pomeroy, J. (2006). ADHD symptom subtypes in children with pervasive developmental disorder. *Journal of Autism and Developmental Disorders, 36*(2), 271–283. https://doi.org/10.1007/s10803-005-0060-3

Gadow, K. D., DeVincent, C. J., Pomeroy, J., & Azizian, A. (2004). Psychiatric symptoms in preschool children with PDD and clinic and comparison samples.

Journal of Autism and Developmental Disorders, 34(4), 379–393. https://doi.org/10.1023/B:JADD.0000037415.21458.93

Gadow, K. D., DeVincent, C. J., Pomeroy, J., & Azizian, A. (2005). Comparison of DSM-IV symptoms in elementary school-age children with PDD versus clinic and community samples. *Autism, 9*(4), 392–415. https://doi.org/10.1177/1362361305056079

George, D., & Mallery, P. (2019). *IBM SPSS statistics 26 step by step: A simple guide and reference*: Routledge.

Gillberg, I. C., Helles, A., Billstedt, E., & Gillberg, C. (2016). Boys with asperger syndrome grow up: psychiatric and neurodevelopmental disorders 20 years after initial diagnosis. Journal of Autism and Developmental Disorders, 46(1), 74–82. https://doi.org/10.1007/s10803-015-2544-0

Gjevik, E., Eldevik, S., Fjaeran-Granum, T., & Sponheim, E. (2011). Kiddie-SADS reveals high rates of DSM-IV disorders in children and adolescents with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 41*(6), 761–769. https://doi.org/10.1007/s10803-010-1095-7

Gobrial, E., & Raghavan, R. (2012). Prevalence of anxiety disorder in children and young people with intellectual disabilities and autism. *Advances in Mental Health and Intellectual Disabilities, 6*(3), 130–140. https://doi.org/10.1108/20441281211227193

Godoy, L. D., Rossignoli, M. T., Delfino-Pereira, P., Garcia-Cairasco, N., & Umeoka, E. H. d. L. (2018). A comprehensive overview on stress neurobiology: Basic concepts and clinical implications. *Frontiers in Behavioral Neuroscience, 12*, 127–127. https://doi.org/10.3389/fnbeh.2018.00127

Gotham, K., Bishop, S. L., Hus, V., Huerta, M., Lund, S., Buja, A., Krieger, A., & Lord, C. (2013). Exploring the relationship between anxiety and insistence on sameness in autism spectrum disorders. *Autism Research, 6*(1), 33–41. https://doi.org/10.1002/aur.1263

Gotham, K., Brunwasser, S. M., & Lord, C. (2015). Depressive and anxiety symptom trajectories from school age through young adulthood in samples with autism spectrum disorder and developmental delay. *Journal of the American Academy of Child & Adolescent Psychiatry, 54*(5), 369–379. https://doi.org/10.1016/j.jaac.2015.02.005

Gray, J. A. (1985). The neuropsychology of anxiety. *Issues in Mental Health Nursing, 7*(1-4), 201–228.

Green, J., Gilchrist, A., Burton, D., & Cox, A. (2000). Social and psychiatric functioning in adolescents with Asperger syndrome compared with conduct disorder. *Journal of Autism and Developmental Disorders, 30*(4), 279–293. https://doi.org/10.1023/A:1005523232106

Green, S. A., & Ben-Sasson, A. (2010). Anxiety disorders and sensory over-responsivity in children with autism spectrum disorders: Is there a causal relationship? *Journal of Autism and Developmental Disorders, 40*(12), 1495–1504. https://doi.org/10.1007/s10803-010-1007-x

Green, S. A., Ben-Sasson, A., Soto, T. W., & Carter, A. S. (2012). Anxiety and sensory over-responsivity in toddlers with autism spectrum disorders: Bidirectional effects across time. *Journal of Autism and Developmental Disorders, 42*, 1112–1119. https://doi.org/10.1007/s10803-011-1361-3

Groden, J., Baron, M. G., & Groden, G. (2006). Assessment and coping strategies. In M. G. Baron, J. Groden, G. Grode, & L. P. Lipsitt (Eds.), *Stress and coping in autism* (pp. 15–41). Oxford University Press.

Grondhuis, S. N., & Aman, M. G. (2012). Assessment of anxiety in children and adolescents with autism spectrum disorders. *Research in Autism Spectrum Disorders, 6*(4), 1345-1365. https://doi.org/10.1016/j.rasd.2012.04.006

Harrell, F. E. J. (2001). *Regression modeling strategies*. Springer-Verlag.

Harrison, P., & Oakland, T. (2003). *Adaptive Behaviour Assessment System user's manual*. Psychological Corporation.

Hassan, M. M., & Mokhtar, H. M. O. (2019). Investigating autism etiology and heterogeneity by decision tree algorithm. *Informatics in Medicine Unlocked, 16*, 100215. https://doi.org/10.1016/j.imu.2019.100215

Helverschou, S. B., Bakken, T. L., & Martinsen, H. (2009). The Psychopathology in Autism Checklist (PAC): A pilot study. *Research in Autism Spectrum Disorders, 3*, 179-195. https://doi.org/10.1016/j.rasd.2008.05.004

Helverschou, S. B., & Martinsen, H. (2011). Anxiety in people diagnosed with autism and intellectual disability: Recognition and phenomenology. *Research in Autism Spectrum Disorders, 5*, 377–387. https://doi.org/10.1016/j.rasd.2010.05.003

Hepburn, S. L., Blakeley-Smith, A., Wolff, B., & Reaven, J. A. (2016). Telehealth delivery of cognitive-behavioral intervention to youth with autism spectrum disorder and anxiety: A pilot study. *Autism: The International Journal of Research and Practice, 20*(2), 207–218. https://doi.org/10.1177/1362361315575164

Hess, K. L. (2009). *Stress for individuals with autism spectrum disorders: Effects of age, gender, and intelligence quotient* [Doctoral thesis, Georgia State University]. ProQuest Dissertations Publishing. https://go.exlibris.link/PBcGbCL8(Dissertation/Thesis)

Hill, J., & Furniss, F. (2006). Patterns of emotional and behavioural disturbance associated with autistic traits in young people with severe intellectual disabilities and challenging behaviours. *Research in Developmental Disabilities, 27*(5), 517–528. https://doi.org/10.1016/j.ridd.2005.07.001

Hollocks, M. J., Howlin, P., Papadopoulos, A. S., Khondoker, M., & Simonoff, E. (2014). Differences in HPA-axis and heart rate responsiveness to psychosocial stress in children with autism spectrum disorders with and without co-morbid anxiety. *Psychoneuroendocrinology, 46*, 32–45.

Hollocks, M. J., Lerh, J. W., Magiati, I., Meiser-Stedman, R., & Brugha, T. S. (2019). Anxiety and depression in adults with autism spectrum disorder: A systematic review and meta-analysis. *Psychological Medicine, 49*(4), 559–572. https://doi.org/10.1017/S0033291718002283

Honey, E., McConachie, H., Turner, M., & Rodgers, J. (2012). Validation of the repetitive behaviour questionnaire for use with children with autism spectrum disorders. *Research in Autism Spectrum Disorders, 6*, 355–364. https://doi.org/10.1016/j.rasd.2011.06.009

Hunsche, M., Cervin, M., Storch, E. A., Kendall, P. C., Wood, J., & Kerns, C. (2022). Social functioning and the presentation of anxiety in children on the autism spectrum: A multimethod, multiinformant analysis. *Journal of Psychopathology and Clinical Science, 131*(2), 198–208. https://doi.org/10.1037/abn0000724

Jahromi, L. B., Meek, S. E., & Ober-Reynoldsa, S. (2012). Emotion regualtion in the context of frustration in children with high functioning autism and their typical peers. *Journal of Child Psychology and Psychiatry, 53*(12), 1250–1258. https://doi.org/10.1111/j.1469-7610.2012.02560.x

Jenkinson, R., Milne, E., & Thompson, A. (2020). The relationship between intolerance of uncertainty and anxiety in autism: A systematic literature review and meta-analysis. *Autism, 24*(8), 1933–1944. https://doi.org/10.1177/1362361320932437

Jiujias, M., Kelley, E., & Hall, L. (2017). Restricted, repetitive behaviors in autism spectrum disorder and obsessive–compulsive disorder: A comparative review. *Child Psychiatry and Human Development, 48*(6), 944–959. https://doi.org/10.1007/s10578-017-0717-0

Joosten, A. V., & Bundy, A. C. (2010). Sensory processing and stereotypical and repetitive behaviour in children with autism and intellectual disability. *Australian Occupational Therapy Journal, 57*(6), 366–372. https://doi.org/10.1111/j.1440-1630.2009.00835.x

Joosten, A. V., Bundy, A. C., & Einfeld, S. L. (2009). Intrinsic and extrinsic motivation for stereotypic and repetitive behavior. *Journal of Autism and Developmental Disorders, 39*(3), 521–531. https://doi.org/10.1007/s10803-008-0654-7

Joosten, A. V., Bundy, A. C., & Einfeld, S. L. (2012). Context influences the motivation for stereotypic and repetitive behaviour in children diagnosed with intellectual disability with and without autism. *Journal of Applied Research in Intellectual Disabilities, 25*(3), 262–270. https://doi.org/10.1111/j.1468-3148.2011.00663.x

Joyce, C., Honey, E., Leekam, S. R., Barrett, S. L., & Rodgers, J. (2017). Anxiety, intolerance of uncertainty and restricted and repetitive behaviour: Insights directly from young people with ASD. *Journal of Autism and Developmental Disorders, 47*(12), 3789–3802. https://doi.org/10.1007/s10803-017-3027-2

Kagan, J., Reznick, S., & Snidman, N. (1988). Biological bases of childhood shyness. *Science, 240*(4849), 167. http://search.proquest.com.libraryproxy.griffith.edu.au/docview/213531422?accountid=14543

Kamphaus, R. W., Huberty, C. J., DiStefano, C., & Petoskey, M. D. (1997). A typology of teacher-rated child behavior for a national U.S. sample. *Journal of Abnormal Child Psychology, 25*(6), 453–463. https://doi.org/10.1023/A:1022681630818

Kamphaus, R. W., Petoskey, M. D., Cody, A. H., Rowe, E. W., Huberty, C. J., & Reynolds, C. R. (1999). A typology of parent rated child behavior for a national U.S. sample. *Journal of Child Psychology and Psychiatry, 40*(4), 607–616. https://doi.org/10.1111/1469-7610.00477

Kanne, S. M., Abbacchi, A. M., & Constantino, J. N. (2009). Multi-Informant ratings of psychiatric symptom severity in children with autism spectrum disorders: The importance of environmental context. *Journal of Autism and Developmental Disorders, 39*(6), 856–864. https://doi.org/10.1007/s10803-009-0694-7

Kanner, L. (1943). Autistic disturbances of affective contact. *Nervous Child, 2*, 217–250.

Kelly, A. B., Garnett, M. S., Attwood, T., & Peterson, C. (2008). Autism spectrum symptomatology in children: The impact of family and peer relationships. *Journal of Abnormal Child Psychology, 36*(7), 1069–1081. https://doi.org/10.1007/s10802-008-9234-8

Kerns, C. M., & Kendall, P. C. (2013). The presentation and classification of anxiety in autism spectrum disorder. *Clinical Psychology: Science and Practice, 19*(4), 323–347. https://doi.org/10.1111/cpsp.12009

Kerns, C. M., Kendall, P. C., Berry, L., Souders, M. C., Franklin, M. E., Shultz, R. T., Miller, J., & Herrington, J. (2014). Traditional and atypical presentations of anxiety in youth with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 44*, 2851–2861. https://doi.org/10.1007/s10803-014-2141-7

Kerns, C. M., Renno, P., Kendall, P. C., Wood, J., & Storch, E. A. (2017). Anxiety Disorders Interview Schedule-Autism Addendum: Reliability and validity in children with autism spectrum disorder. *Journal of Clinical Child and*

Adolescent Psychology, 46(1), 88–100. https://doi.org/10.1080/15374416.2016.1233501

Kildahl, A. N., Bakken, T. L., Matre, E. A. W., Hellerud, J. M. A., Engebretsen, M. H., & Helverschou, S. B. (2021). Case study: Identification of anxiety and subsequent intervention in an adolescent male with autism, severe intellectual disability and self-injurious behaviour. *International Journal of Developmental Disabilities, 67*(5), 327–338. https://doi.org/10.1080/20473869.2020.1850160

Kim, J. Y., & Ha, E. H. (2020). Cluster analysis of the Child Behavior Checklist 1.5–5 for preschool children diagnosed with a mental disorder. *Psychological Reports, 123*(4), 1403–1424. https://doi.org/10.1177/0033294119844980

Kim, Y. S., Leventhal, B. L., Koh, Y.-J., Fombonne, E., Laska, E., Lim, E.-C., Cheon, K.-A., Kim, S.-J., Kim, Y.-K., Lee, H., Song, D.-H., & Grinker, R. R. (2011). Prevalence of autism spectrum disorders in a total population sample. *The American Journal of Psychiatry, 168*(9), 904–912. https://doi.org/10.1176/appi.ajp.2011.10101532

Knapp, M., Romeo, R., & Beecham, J. (2009). Economic cost of autism in the uk. *Autism: The International Journal of Research and Practice, 13(3),* 317–336. https://doi.org/10.1177/1362361309104246

Kolacz, J., Raspa, M., Heilman, K. J., & Porges, S. W. (2018). Evaluating sensory processing in Fragile X syndrome: Psychometric analysis of the Brain Body Center Sensory Scales (BBCSS). *Journal of Autism and Developmental Disorders, 48*(6), 2187–2202. https://doi.org/10.1007/s10803-018-3491-3

Koritsas, S., & Iacono, T. (2015). Predictors of challenging behaviour in adults with intellectual disability. *Advances in Mental Health and Intellectual Disabilities, 9*(6), 312–326. https://doi.org/10.1108/AMHID-06-2015-0029

Lai, M.-C., Kassee, C., Besney, R., Bonato, S., Hull, L., Mandy, W., Szatmari, P., & Ameis, S. H. (2019). Prevalence of co-occurring mental health diagnoses in the autism population: A systematic review and meta-analysis. *The Lancet Psychiatry, 6*(10), 819–829. https://doi.org/10.1016/S2215-0366(19)30289-5

Lainhart, J. E. (1999). Psychiatric problems in individuals with autism, their parents and siblings. *International Review of Psychiatry, 11*(4), 278–298. https://doi.org/10.1080/09540269974177

Lainhart, J. E., & Folstein, S. E. (1994). Affective disorders in people with autism: A review of published cases. *Journal of Autism & Developmental Disorders, 24*(5), 587–601.

Lecavalier, L. (2006). Behavioral and emotional problems in young people with pervasive developmental disorders: Relative prevalence, effects of subject characteristics and empirical classification. *Journal of Autism and Developmental Disorders, 36*(8), 1101–1114. https://doi.org/10.1007/s10803-006-0147-5

Lecavalier, L., Aman, M. G., Hammer, D., Stoica, W., & Mathews, G. L. (2004). Factor analysis of the Nisonger Child Behavior Rating Form in children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 34*(6), 709–721. https://doi.org/10.1007/s10803-004-5291-1

Leekam, S. R., Prior, M. R., & Uljarevic, M. (2011). Restricted and repetitive behaviours in autism spectrum disorders: A review of research in the last decade. *Psychological Bulletin, 137*(4), 562–593. https://doi.org/10.1037/a0023341

Leekam, S., Tandos, J., McConachie, H., Meins, E., Parkinson, K., Wright, C., Turner, M., Arnott, B., Vittorini, L., & Le Couteur, A. (2007). Repetitive behaviours in typically developing 2-year-olds. *The Journal of Child Psychology and*

Psychiatry, 48(11), 1131–1138. https://doi.org/10.1111/j.1469-7610.2007.01778.x

Leslie, A. M. (1987). Pretense and representation: The origins of "theory of mind". *Psychological Review, 94*(4), 412–426. https://doi.org/10.1037/0033-295X.94.4.412

Leyfer, O. T., Folstein, S. E., Bacalman, S., Davis, N. O., Dinh, E., Morgan, J., Tager-Flusberg, H., & Lainhart, J. E. (2006). Comorbid psychiatric disorders in children with autism: Interview development and rates of disorders. *Journal of Autism and Developmental Disorders, 36*(7), 849–861. https://doi.org/10.1007/s10803-006-0123-0

Liss, M., Mailoux, J., & Erchull, M. J. (2008). The relationships between sensory processing sensitivity, alexithymia, autism, depression and anxiety. *Personality and Individual Differences*(45), 255–259. https://doi.org/10.1016/j.paid.2008.04.009

Llanes, E., Blacher, J., Stavropoulos, K., & Eisenhower, A. (2018). Parent and teacher reports of comorbid anxiety and ADHD symptoms in children with ASD. *Journal of Autism and Developmental Disorders, 50*(5), 1520–1531. https://doi.org/10.1007/s10803-018-3701-z

Lord, C., Pickles, A., McLennan, J., Rutter, M., Bregman, J., Folstein, S., Fombonne, E., Leboyer, M., & Minshew, N. (1997). Diagnosing autism: Analyses of data from the Autism Diagnostic Interview. *Journal of Autism & Developmental Disorders, 27*, 501–517.

MacNeil, B. M., Lopes, V. A., & Minnes, P. M. (2009). Anxiety in children and adolescents with autism spectrum disorders. *Research in Autism Spectrum Disorders, 3*, 1–21. https://doi.org/10.1016/j.rasd.2008.06.001

Matson, J. L., & Cervantes, P. E. (2014). Commonly studied comorbid psychopathologies among persons with autism spectrum disorder. *Research in Developmental Disabilities, 35*(5), 952–962. https://doi.org/10.1016/j.ridd.2014.02.012

Matson, J. L., & Nebel-Schwalm, M. S. (2007). Comorbid psychopathology with autism spectrum disorder in children: An overview. *Research in Developmental Disabilities, 28*(4), 341–352. https://doi.org/10.1016/j.ridd.2005.12.004

Matson, J. L., & Shoemaker, M. (2009). Intellectual disability and its relationship to autism spectrum disorders. *Research in Developmental Disabilities, 30*(6), 1107–1114. https://doi.org/10.1016/j.ridd.2009.06.003

Matson, J. L., Wilkins, J., & Macken, J. (2008). The relationship of challenging behaviours to severity and symptoms of autism spectrum disorders. *Journal of Mental Health Research in Intellectual Disabilities, 2*(1), 29–44. https://doi.org/10.1080/19315860802611415

May, T., Brignell, A., & Williams, K. (2020). Autism spectrum disorder prevalence in children aged 12–13 years from the Longitudinal Study of Australian Children. *Autism Research, 13*(5), 821–827. https://doi.org/10.1002/aur.2286

Mayes, S. D., Calhoun, S. L., Murray, M. J., Ahuja, M., & Smith, L. A. (2011). Anxiety, depression, and irritability in children with autism relative to other neuropsychiatric disorders and typical development. *Research in Autism Spectrum Disorders, 5*(1), 474–485. https://doi.org/10.1016/j.rasd.2010.06.012

McCarthy, J., Hemmings, C., Kravariti, E., Dworzynski, K., Holt, G., Bouras, N., & Tsakanikos, E. (2010). Challenging behaviour and co-morbid psychopathology in adults with intellectual disability and autism spectrum disorders. *Research in Develomental Disabilities, 31*, 362–366. https://doi.org/10.1016/j.ridd.2009.10.009

McConachie, H., McLaughlin, E., Grahame, V., Taylor, H., Honey, E., Tavernor, L., Rodgers, J., Freeston, M., Hemm, C., Steen, N., & Le Couteur, A. (2014). Group therapy for anxiety in children with autism spectrum disorder. *Autism: The International Journal of Research and Practice, 18*(6), 723–-732. https://doi.org/10.1177/1362361313488839

McTiernan, A., Leader, G., Healy, O., & Mannion, A. (2011). Analysis of risk factors and early predictors of challenging behaviour for children with autism spectrum disorder. *Research in Autism Spectrum Disorders, 5*, 1215–1222. https://doi.org/10.1016/j.rasd.2011.01.009

Militerni, R., Bravaccio, C., Falco, C., Fico, C., & Palermo, M. T. (2002). Repetitive behaviours in autistic disorder. *European Child and Adolescent Psychiatry, 11*, 210–218. https://doi.org/10.1007/s00787-002-0279-x

Mineka, S., & Zinbarg, R. (2006). A contemporary learning theory perspective on the etiology of anxiety disorders: It's not what you thought it was. *The American Psychologist, 61*(1), 10–26. https://doi.org/10.1037/0003-066X.61.1.10

Mingins, J. E., Tarver, J., Waite, J., Jones, C., & Surtees, A. D. R. (2021). Anxiety and intellectual functioning in autistic children: A systematic review and meta-analysis. *Autism, 25*(1), 18–32. https://doi.org/10.1177/1362361320953253

Mogg, K., & Bradley, B. P. (1998). A cognitive-motivational analysis of anxiety. *Behaviour Research and Therapy, 36*(9), 809-848. https://doi.org/10.1016/S0005-7967(98)00063-1

Mohapatra, S., Jahan, M., & Kishore, M. T. (2017). Autistic spectrum disorders in children with intellectual disability. *Indian Journal of Clinical Psychology, 45*(1), 35–40.

Morgan, K. (2006). Is autism a stress disorder? What studies of nonautistic populations can tell us. In M. G. Baron, J. Groden, G. Groden, & L. P. Lipsitt (Eds.), *Stress and coping in autism* (pp. 129–182). Oxford University Press.

Moskowitz, L. J., Mulder, E., Walsh, C. E., McLaughlin, D. M., Zarcone, J. R., Proudfit, G. H., & Carr, E. G. (2013). A multimethod assessment of anxiety and problem behavior in children with autism spectrum disorders and intellectual disability. *American Journal on Intellectual and Developmental Disabilities, 118*(6), 419–434. https://doi.org/10.1352/1944.7558.118.6.419

Moskowitz, L. J., Rosen, T., Lerner, M. D., & Levine, K. (2017). Chapter 5 - Assessment of anxiety in youth with autism spectrum disorder. In C. M. Kerns, P. Renno, E. A. Storch, P. C. Kendall, & J. J. Wood (Eds.), *Anxiety in children and adolescents with autism spectrum disorder* (pp. 79–104). Academic Press.

Moskowitz, L. J., Walsh, C. E., Mulder, E., McLaughlin, D. M., Hajcak, G., Carr, E. G., & Zarcone, J. R. (2017). Intervention for anxiety and problem behavior in children with autism spectrum disorder and intellectual disability. *Journal of Autism and Developmental Disorders, 47*(12), 3930–3948. https://doi.org/10.1007/s10803-017-3070-z

Muris, P., Steerneman, P., Merckelbach, H., Holdrinet, I., & Meesters, C. (1998). Comorbid anxiety symptoms in children with pervasive developmental disorders. *Journal of Anxiety Disorders, 12*(4), 387–393. https://doi.org/10.1016/S0887-6185(98)00022-X

Murphy, O., Healy, O., & Leader, G. (2009). Risk factors for challenging behaviors among 157 children with autism spectrum disorder in Ireland. *Research in Autism Spectrum Disorders, 3*, 474–482. https://doi.org/10.1016/j.rasd.2008.09.008

Myers, S. M., Voigt, R. G., Colligan, R. C., Weaver, A. L., Storlie, C. B., Stoeckel, R. E., Port, J. D., & Katusic, S. K. (2018). Autism spectrum disorder: Incidence and time trends over two decades in a population-based birth cohort. *Journal of Autism and Developmental Disorders, 49*(4), 1455–1474. https://doi.org/10.1007/s10803-018-3834-0

Nicholls, G., Hastings, R. P., & Grindle, C. (2020). Prevalence and correlates of challenging behaviour in children and young people in a special school setting. *European Journal of Special Needs Education, 35*(1), 40–54. https://doi.org/10.1080/08856257.2019.1607659

Niditch, L. A., Varela, R. E., Kamps, J. L., & Hill, T. (2012). Exploring the association between cognitive functioning and anxiety in children with autism spectrum disorders: The role of social understanding and aggression. *Journal of Clinical Child and Adolescent Psychology, 41*(2), 127–137. https://doi.org/10.1080/15374416.2012.651994

Nimmo-Smith, V., Heuvelman, H., Dalman, C., Lundberg, M., Idring, S., Carpenter, P., Magnusson, C., & Rai, D. (2020). Anxiety disorders in adults with autism spectrum disorder: A population-based study. *Journal of Autism and Developmental Disorders, 50*(1), 308–318. https://doi.org/10.1007/s10803-019-04234-3

Norris, M., & Lecavalier, L. (2011). Evaluating the validity of the Nisonger Child Behavior Rating Form – Parent Version. *Research in Developmental Disabilities, 32*(6), 2894–2900. https://doi.org/10.1016/j.ridd.2011.05.015

Oliver, C., Licence, L., & Richards, C. (2017). Self-injurious behaviour in people with intellectual disability and autism spectrum disorder. *Current Opinion in Psychiatry, 30*(2), 97–101. https://doi.org/10.1097/YCO.0000000000000313

Park, S. H., Song, Y. J. C., Demetriou, E. A., Pepper, K. L., Thomas, E. E., Hickie, I. B., & Guastella, A. J. (2020). Validation of the 21-item Depression, Anxiety, and Stress Scales (DASS-21) in individuals with autism spectrum disorder. *Psychiatry Research, 291.* https://doi.org/10.1016/j.psychres.2020.113300

Perkins-Dock, R. (2003). Adaptive behaviour assessment system. *Rehabilitation Counselling Bulletin, 46*(3), 183–186.

Podlesnik, C. A., & Kelley, M. E. (2017). Beyond intervention: Shaping policy for addressing persistence and relapse of severe problem behavior. *Policy Insights From the Behavioral and Brain Sciences, 4*(1), 17–24. https://doi.org/10.1177/2372732216683403

Prizant, B. M. (1996). Brief report: Communication, language, social, and emotional development. *Journal of Autism and Developmental Disorders, 26*(2), 173–178. https://doi.org/10.1007/BF02172007

Prizant, B., Wetherby, A., Rubin, E., & Laurent, A. (2003). The SCERTS model: A transactional, family-centred approach to enhancing communications and socioemotional abilities of children with autism spectrum disorder. *Infants and Young Children, 16*(4), 296–316.

Pruijssers, A. C., van Meijel, B., Maaskant, M., Nijssen, W., & van Achterberg, T. (2014). The relationship between challenging behaviour and anxiety in adults with intellectual disabilities: A literature review. *Journal of Intellectual Disability Research, 58*(2), 162–171. https://doi.org/10.1111/jir.12012

Rachman, S. (1977). The conditioning theory of fear-acquisition: A critical examination. *Behaviour Research and Therapy, 15*(5), 375–387. https://doi.org/10.1016/0005-7967(77)90041-9

Raftery, A. E. (1995). Bayesian model selection in social research. *Sociological Methodology, 25*, 111–163. https://doi.org/10.2307/271063

Raftery, A. E., Madigan, D., & Hoeting, J. A. (1997). Bayesian model averaging for linear regression models. *Journal of the American Statistical Association, 92*(437), 179–191. https://doi.org/10.1080/01621459.1997.10473615

Rajendran, G., & Mitchell, P. (2007). Cognitive theories of autism. *Developmental Review, 27*, 224–260. https://doi.org/10.1016/j.d.r.2007.02.001

Ranciati, S., Galimberti, G., & Soffritti, G. (2018). Bayesian variable selection in linear regression models with non-normal errors. *Statistical Methods & Applications, 28*(2), 323–358. https://doi.org/10.1007/s10260-018-00441-x

Reardon, T. C., Gray, K. M., & Melvin, G. A. (2015). Anxiety disorders in children and adolescents with intellectual disability: Prevalence and assessment. *Research in Developmental Disabilities, 36*, 175-190. https://doi.org/10.1016/j.ridd.2014.10.007

Richler, J., Huerta, M., Bishop, S. L., & Lord, C. (2010). Developmental trajectories of restricted and repetitive behaviors and interests in children with autism spectrum disorders. *Development and Psychopathology, 22*(1), 55–69. https://doi.org/10.1017/S0954579409990265

Rivard, M., Terroux, A., Mercier, C., & Parent-Boursier, C. (2015). Indicators of intellectual disabilities in young children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 45*, 127–137. https://doi.org/10.1007/s10803-014-2198-3

Rodgers, J., Glod, M., Connolly, B., & McConachie, H. (2012). The relationship between anxiety and repetitive behaviours in autism spectrum disorder. *Journal of Autism and Developmental Disorders, 42*(11), 2404–2409. https://doi.org/10.1007/s10803-012-1531-y

Rodgers, J., Hodgson, A., Shields, K., Wright, C., Honey, E., & Freeston, M. (2016). Towards a treatment for intolerance of uncertainty in young people with autism spectrum disorder: Development of the Coping with Uncertainty in Everyday

Situations (CUES©) programme. *Journal of Autism and Developmental Disorders, 47*(12), 3959–3966. https://doi.org/10.1007/s10803-016-2924-0

Rodgers, J., Wigham, S., McConachie, H., Freeston, M., Honey, E., & Parr, J. R. (2016). Development of the Anxiety Scale for Children with Autism Spectrum Disorder (ASC-ASD). *Autism Research, 9*(11), 1205–1215. https://doi.org/10.1002/aur.1603

Romanczyk, R. G., & Gillis, J. M. (2006). Autism and the physiology of stress and anxiety. In M. G. Baron, J. Groden, G. Groden, & L. P. Lipsitt (Eds.), *Stress and coping in autism* (pp. 183–204). Oxford University Press.

Russell, K. M., Frost, K. M., & Ingersoll, B. (2019). The relationship between subtypes of repetitive behaviors and anxiety in children with autism spectrum disorder. *Research in Autism Spectrum Disorders, 62*, 48–54. https://doi.org/10.1016/j.rasd.2019.03.006

Rutter, M., Bailey, A., & Lord, C. (2003). *Social Communication Questionnaire (SCQ)*. Western Psychological Services.

Rzepecka, H., McKenzie, K., McClure, I., & Murphy, S. (2011). Sleep, anxiety and challenging behaviour in children with intellectual disability and/or autism spectrum disorder. *Research in Developmental Disabilities, 32*(6), 2758–2766. https://doi.org/10.1016/j.ridd.2011.05.034

Samson, A. C., Hardan, A. Y., Lee, I. A., Phillips, J. M., & Gross, J. J. (2015). Maladaptive behavior in autism spectrum disorder: The role of emotion experience and emotion regulation. *Journal of Autism and Developmental Disorders, 45*(11), 3424–3432. https://doi.org/10.1007/s10803-015-2388-7

Samson, A. C., Phillips, J. M., Parker, K. J., Shah, S., Gross, J. J., & Hardan, A. Y. (2014). Emotion dysregulation and the core features of autism spectrum disorder. *Journal of Autism and Developmental Disorders, 44*(7), 1766–1772. https://doi.org/10.1007/s10803-013-2022-5

Silverman, W. K., & Albano, A. M. (1996). *The Anxiety Disorders Interview Schedule for Children (ADIS-C/P)*. Psychological Corporation.

Simonoff, E., Pickles, A., Charman, T., Chandler, S., Loucas, T., & Baird, G. (2008). Psychiatric disorders in children with autism spectrum disorders: Prevalence, comorbidity, and associated factors in a population-derived sample. *Journal of the American Academy of Child and Adolescent Psychiatry, 47*(8), 921–929. https://doi.org/10.1097/CHI.0b013e318179964f

Sims, M., Guilfoyle, A., & Parry, T. S. (2006). Children's cortisol levels and quality of child care provision. *Child: Care, Health and Development, 32*(4), 453–466. https://doi.org/10.1111/j.1365-2214.2006.00632.x

Smiley, K., Gerstein, B., & Nelson, S. (2018). Unveiling the autism epidemic. *Journal of Neurology and Clinical Neuroscience, 2*(2), 1.

Sofronoff, K., Attwood, T., & Hinton, S. (2005). A randomised controlled trial of a CBT intervention for anxiety in children with Asperger's syndrome. *Child Psychology and Psychiatry, 46*(11), 1152-1160. https://doi.org/10.1111/j.1469–7610.2005.00411.x

South, M., Ozonoff, S., & McMahon, W. M. (2005). Repetitive behavior profiles in Asperger syndrome and high-functioning autism. *Journal of Autism and Developmental Disorders, 35*(2), 145–158. https://doi.org/10.1007/s10803-004-1992-8

South, M., & Rodgers, J. (2017). Sensory, emotional and cognitive contributions to anxiety in autism spectrum disorders. *Frontiers in Human Neuroscience, 11*(20). https://doi.org/10.3389/fnhum.2017.00020

Spiker, M. A., Lin, C. E., Van Dyke, M., & Wood, J. J. (2011). Restricted interests and anxiety in children with autism. *Autism, 16*(3), 306–320. https://doi.org/10.1177/1362361311401763

Sukhodolsky, D. G., Scahill, L., Gadow, K. D., Arnold, L. E., Aman, M. G., McDougle, C. J., McCracken, J. T., Tierney, E., White, S. W., Lecavalier, L., & Vitiello, B. (2008). Parent-Rated anxiety symptoms in children with pervasive developmental disorders: Frequency and association with core autism symptoms and cognitive functioning. *Journal of Abnormal Child Psychology, 36*(1), 117–128. https://doi.org/10.1007/s10802-007-9165-9

Sweeten, T. L., Posey, D. J., Shekhar, A., & McDougle, C. J. (2002). The amygdala and related structures in the pathophysiology of autism. *Pharmacology Biochemistry and Behavior, 71*(3), 449–455. https://doi.org/10.1016/S0091-3057(01)00697-9

Syu, Y.-C., Huang, P.-C., Wang, T.-Y., Chang, Y.-C., & Lin, L.-Y. (2020). Relationship among sensory over-responsivity, problem behaviors, and anxiety in emerging adults with autism spectrum disorder. *Neuropsychiatric Disease and Treatment, 16*, 2181–2190. https://doi.org/10.2147/NDT.S270308

Tabachnick, B., & Fidell, L. (2019). *Using multivariate statistics* (7th ed.). Pearson.

Taffe, J. R., Tonge, B. J., Gray, K., M., & Einfield, S. L. (2008). Extracting more information from behaviour checklists by using components of mean based scores. *International Journal of Methods in Psychiatric Research, 17*(4), 232–240. https://doi.org/10.1002/mpr.260

Tantam, D. (2000). Psychological disorder in adolescents and adults with Asperger syndrome. *Autism, 4*(1), 47–62. https://doi.org/10.1177/1362361300004001004

Tassé, M. J., Aman, M. G., Hammer, D., & Rojahn, J. (1996). The Nisonger child behavior rating form: Age and gender effects and norms. *Research in*

Developmental Disabilities, 17(1), 59–75. https://doi.org/10.1016/0891-4222(95)00037-2

Toscano, R., Hudson, J. L., Baillie, A. J., Lyneham, H. J., & McLellan, L. F. (2020). Development of the Macquarie Anxiety Behavioural Scale (MABS): A parent measure to assess anxiety in children and adolescents including young people with autism spectrum disorder. *Journal of Affective Disorders, 276*, 678–685. https://doi.org/10.1016/j.jad.2020.06.076

Tsai, L. Y. (1996). Brief report: Comorbid psychiatric disorders of autistic disorder. *Journal of Autism and Developmental Disorders, 26*(2), 159–163. https://doi.org/10.1007/BF02172004

Tsai, L. (2006). Diagnosis and treatment of anxiety disorders in individuals with autism spectrum disorder. In M. G. Baron, J. Groden, G. Groden, & L. P. Lipsitt (Eds.), *Stress and coping in autism* (pp. 388–440). Oxford University Press.

Vakhitova, Z. I., & Alston-Knox, C. L. (2018). Non-significant p-values? Strategies to understand and better determine the importance of effects and interactions in logistic regression. *PLoS One, 13*(11). https://doi.org/10.1371/journal.pone.0205076

van Andel, H., Jansen, L., Grietens, H., Knorth, E., & van der Gaag, R. (2014). Salivary cortisol: A possible biomarker in evaluating stress and effects of intervention in young foster children? *European Child and Adolescent Psychiatry, 23*, 3–12. https://doi.org/10.1007/s00787-013-0439-1

Van Ryzin, M., Chatham, M., Kryzer, E., Kertes, D., & Gunnar, M. (2009). Identifying atypical cortisol patterns in young children: The benefits of group-based trajectory modeling. *Psychoneuroendocrinology, 34*, 50–61. https://doi.org/10.1016/j.psyneuen.2008.08.014

Vasa, R. A., Kalb, L., Mazurek, M., Kanne, S., Freedman, B., Keefer, A., Clemons, T., & Murray, D. (2013). Age-related differences in the prevalence and correlates of anxiety in youth with autism spectrum disorders. *Research in Autism Spectrum Disorders, 7*(11), 1358–1369. https://doi.org/10.1016/j.rasd.2013.07.005

Weisbrot, D. M., Gadow, K. D., DeVincent, C. J., & Pomeroy, J. (2005). The presentation of anxiety in children with pervasive developmental disorders. *Journal of Child and Adolescent Psychopharmacology, 15*(3), 477–496. https://doi.org/10.1089/cap.2005.15.477

White, S. W., Oswald, D., Ollendick, T., & Scahill, L. (2009). Anxiety in children and adolescents with autism spectrum disorders. *Clinical Psychology Review, 29*, 216–229. https://doi.org/10.1016/j.cpr.2009.01.003

Wigham, S., Rodgers, J., South, M., McConachie, H., & Freeston, M. (2014). The interplay between sensory processing abnormalities, intolerance of uncertainty, anxiety and restricted and repetitive behaviours in autism spectrum disorder. *Journal of Autism and Developmental Disorders, 45*(4), 943–952. https://doi.org/10.1007/s10803-014-2248-x'

Williams, J., Watts, F., Macleod, C., & Mathews, A. (1997). *Cognitive psychology and emotional disorders.* John Wiley & Sons.

Williams, S., Leader, G., Mannion, A., & Chen, J. (2015). An investigation of anxiety in children and adolescents with autism spectrum disorder. *Research in Autism Spectrum Disorders, 10*, 30–40. https://doi.org/10.1016/j.rasd.2014.10.017

Williams, Z. J., Failla, M. D., Gotham, K. O., Woynaroski, T. G., & Cascio, C. (2018). Psychometric evaluation of the Short Sensory Profile in Youth with Autism Spectrum Disorder. *Journal of Autism and Developmental Disorders, 48*(12), 4231–4249. https://doi.org/10.1007/s10803-018-3678-7

Witwer, A. N., & Lecavalier, L. (2010). Validity of comorbid psychiatric disorders in youngsters with autism spectrum disorder. *Journal of Developmental and Physical Disabilities, 22*(4), 367–380. https://doi.org/10.1007/s10882-010-9194-0

Wood, J. J., & Gadow, K. D. (2010). Exploring the nature and function of anxiety in youth with autism spectrum disorders. *Clinical Psychology: Science and Practice, 17*(4), 281–292. https://doi.org/10.1111/j.1468-2850.2010.01220.x

Yeargin-Allsopp, M., Rice, C., Karapurkar, T., Doernberg, N., Boyle, C., & Murphy, C. (2003). Prevalence of autism in a US metropolitan area. *The Journal of the American Medical Association, 289*(1), 49–55. https://doi.org/10.1001/jama.289.1.49

Zeidan, J., Fombonne, E., Scorah, J., Ibrahim, A., Durkin, M. S., Saxena, S., Yusuf, A., Shih, A., & Elsabbagh, M. (2022). Global prevalence of autism: A systematic review update. *Autism Research, 15*(5), 778–790. https://doi.org/10.1002/aur.2696

Appendix A

Griffith University Human Research Ethics Approval

Full Research Ethics Clearance 2016/010

RG

RIMS Griffith [redacted]

Friday, 5 February 2016 at 9:51 am

To: Helen Appleton; Trevor Clark; Allison Waters; Jacqueline Roberts; **Cc:** research-ethics; Kim Madison

This message is high priority.

GRIFFITH UNIVERSITY HUMAN RESEARCH ETHICS REVIEW

Dear Prof Jacqueline Roberts

I write further to the additional information provided in relation to the provisional approval granted to your application for ethical clearance for your project "An exploration of the presence and behavioural manifestation of anxiety in children and young actuals with autism spectrum disorder and intellectual disability" (GU Ref No: 2016/010).

This is to confirm that this response has addressed the comments and concerns of the HREC.

The ethics reviewers resolved to grant your application a clearance status of "Fully Approved".

Consequently, you are authorised to immediately commence this research on this basis.

Regards

Kim Madison
Policy Officer, Human Research Ethics and Integrity
Office for Research
Bray Centre, Nathan Campus
Griffith University
[redacted]

Appendix B

Giant Steps Ethics Approval

...imagine...include...innovate

Giant Steps Sydney Ltd

3 February 2016

Re: Research Project: *"An exploration of the presence and behavioural manifestation of anxiety in children and young adults with autism spectrum disorder and intellectual disability"*

Giant Steps approves the researchers Professor Jacqui Roberts and Helen Appleton to have access to the families who attend Giant Steps for the purposes of recruitment into this research study.

This includes families who have children currently enrolled in our preschool, school and community college, and could be extended to the alumni family network if required.

Access includes Parent Information Evenings, Friday Footnotes (our weekly newsletter) and the availability to email and mail outs to all parents.

The outcomes of this research promise to be beneficial not only to the families whose children attend Giant Steps, but to the wider community, families and professionals working with individuals with ASD and ID.

We look forward to seeing the results of this research

Kerrie Nelson
School Principal
Giant Steps

Giant Steps Sydney Ltd
ABN 51 087 759 984

"Step House"
23 Punt Road
Gladesville NSW 2111

PO Box 209
Gladesville NSW 1675

[redacted]

www.giantsteps.net.au