THE DEVELOPMENT OF ANXIETY IN CHILDREN WITH AUTISM SPECTRUM DISORDERS

by

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Abstract

Although up to 40% of children with autism spectrum disorder (ASD) have a comorbid anxiety disorder, little is known about the origins and trajectory of change in anxiety symptoms in ASD. Characteristics specific to ASD such as social impairments and alexithymia may alter the experience of anxiety in this population. Consequently, anxiety may differ in the ASD population and merits focused study. This dissertation consists of two related studies that used data from the longitudinal Pathways in ASD study. The psychometric properties of the Spence Children’s Anxiety Scale – Parent Form (SCAS-P) in 238 children who were seen annually from ages 7.5 to 11 were examined in Study 1. While the original six-factor structure was not a good fit in this sample, four subscales reflecting Generalized, Separation Anxiety, Panic and Agoraphobia symptoms were identified. In Study 2, parent ratings of Generalized, Separation Anxiety, Panic and Agoraphobia symptoms were captured at snapshots in middle childhood, as well as changing over time in 262 children who were seen annually between ages 7.5 to 11. The proportion of children whose parents rated them as experiencing Elevated Generalized Anxiety was comparable to past reports, though rates of Elevated Separation Anxiety symptoms were higher than past reports. Parent-rated Generalized Anxiety, Separation Anxiety, Panic and Agoraphobia symptoms were stable over the middle childhood years, and there was little variance in the trajectories of all except the Separation Anxiety domain. Children with age-typical language abilities were rated as experiencing higher levels of Generalized and Separation Anxiety in middle childhood. Parent-rated anxiety in early childhood significantly predicted higher Generalized and Separation anxiety across middle childhood, while parental internalizing symptoms in early childhood were predictive of Generalized, but not Separation Anxiety symptoms. There were no differences in Generalized or Separation Anxiety levels across ages 7-11 between boys and girls. The results of this research offer a deeper understanding of the
psychometric properties of one widely used anxiety rating scale, as well as its predictors, incidence and development over middle childhood. In turn, this understanding can support efforts aimed at preventing and treating anxiety disorders in ASD.


**Lay Abstract**

Children with Autism Spectrum Disorder are at a higher risk of developing an anxiety disorder than children in the general population. However, anxiety in children with autism is not well understood. Study 1 of this dissertation suggested an improved way to measure anxiety in children with autism. Study 2 found that on average, parent’s ratings of Generalized Anxiety, Separation Anxiety, Panic and Agoraphobia levels in children with autism remain stable across middle childhood (ages 7-11), and are higher in children with age-typical language abilities. No differences in Generalized or Separation Anxiety symptoms were found between boys and girls. Several early risk factors for different types of anxiety in middle childhood were identified, including a parent’s own mental health symptoms and anxiety early on in the child’s life. These findings are important for the accurate measurement, treatment and prevention of anxiety disorders in children with autism.
Preface

This dissertation is an original intellectual product of the author, E. Jitlina, who was responsible for all major areas of idea formation, data analysis, and manuscript preparation in Chapters 3 and 4. The study utilized data collected for the “Autism Spectrum Disorders: Pathways to Better Outcomes” research project that was first approved by UBC’s Behavioural Research Ethics Board (BREB) on June 26, 2009 under certificate H09-01085. The Pathways in ASD study team approved use of data for Ms. Jitlina’s thesis on February 19, 2015 and her name was added to BREB on August 10, 2015 (H09-01085-A014). Drs. Ford, Mirenda, and Zumbo offered input and expertise during the formulation of the research, data analysis and manuscript writing. A version of Chapter 3 was published in the Journal of Autism and Developmental Disorders. As per publication guidelines, the co-authors (and principal investigators of the Pathways in ASDs study) offered feedback on the manuscript for submission.
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<td>Attention-deficit hyperactivity disorder</td>
</tr>
<tr>
<td>APA</td>
<td>American Psychiatric Association</td>
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<tr>
<td>ASD</td>
<td>Autism spectrum disorder</td>
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<tr>
<td>CASI</td>
<td>Child and Adolescent Symptom Inventory</td>
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<tr>
<td>CBCL-1.5/5</td>
<td>Child Behavior Checklist for children ages 1.5 to 5 years</td>
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<td>CBCL-6-18</td>
<td>Child Behavior Checklist for children ages 6 to 18 years</td>
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<tr>
<td>CFA</td>
<td>Confirmatory factor analysis</td>
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<td>CFI</td>
<td>Comparative fit index</td>
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<td>DSM-5</td>
<td>Diagnostic and Statistical Manual of Mental Disorders, 5th Edition</td>
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<td>DSM-IV-TR</td>
<td>Diagnostic and Statistical Manual of Mental Disorders, 4th Edition, Text Revision</td>
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<tr>
<td>GAD</td>
<td>Generalized anxiety disorder</td>
</tr>
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<td>LMR</td>
<td>Lifetime morbid risk</td>
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<tr>
<td>MASC-C</td>
<td>Multidimensional Anxiety Scale for Children-Child Report</td>
</tr>
<tr>
<td>MASC-P</td>
<td>Multidimensional Anxiety Scale for Children-Parent Report</td>
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<tr>
<td>MMPI</td>
<td>Minnesota Multiphasic Personality Inventory</td>
</tr>
<tr>
<td>M-P-R</td>
<td>Merrill-Palmer-Revised Scales of Development</td>
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<tr>
<td>OCD</td>
<td>Obsessive compulsive disorder</td>
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<td>RCADS</td>
<td>Revised Child Anxiety and Depression Scale</td>
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<tr>
<td>RMSEA</td>
<td>Root mean square error of approximation</td>
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<tr>
<td>SCARED</td>
<td>Screen for Child Anxiety Related Emotional Disorders</td>
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<td>SCAS-P</td>
<td>Spence Children's Anxiety Scale-Parent form</td>
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<tr>
<td>SCL-90-R</td>
<td>Symptom Checklist-90 Revised</td>
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<td>SRMR</td>
<td>Standardized root mean square residual</td>
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Chapter 1: Introduction

Overview

The last several decades of research on ASD have shown that the core symptoms of ASD rarely occur in isolation. Comorbidity with various medical conditions and psychopathologies is very common (Bauman, 2010; Matson & Nebel-Schwalm, 2007). Anxiety disorders represent one of the most highly prevalent comorbid psychopathologies for individuals with ASD (de Bruin, Ferdinand, Meester, de Nijs, & Verheij, 2006; Leyfer et al., 2006; Simonoff et al., 2008; Skokauskas & Gallagher, 2010) and result in impairment above and beyond that of ASD alone (Ozsivadjian, Knott, & Magiati, 2012). Positively, there is accumulating evidence that anxiety disorders in children with ASD are highly amenable to treatment through cognitive behavioural therapy, with effect sizes that exceed those seen in children without ASD (Danial & Wood, 2013; Sukhodolsky, Bloch, Panza, & Reichow, 2013; Vasa et al., 2014). Examining the early predictors of anxiety disorders in ASD and how these symptoms develop in childhood can encourage preventative efforts, reducing the burden of this comorbidity.

Research in otherwise typically developing individuals suggests that the period from middle childhood through adolescence is a critical developmental period for anxiety disorders. In fact, the risk of developing an anxiety disorder decreases substantially following this high-risk period (Kessler et al., 2005). Moreover, the risk for developing different anxiety disorders varies with age, as suggested by variation in typical ages of onset. Specific phobias and separation anxiety disorder tend to develop in childhood; social anxiety disorder and obsessive compulsive disorder (OCD) often have an onset in early adolescence; and generalized anxiety disorder (GAD), panic disorder, and post-traumatic stress disorder develop in early adulthood (Costello, Egger, & Angold, 2005; Kessler et al., 2005; 2007).
Given the chronic nature of untreated anxiety disorders throughout the lifetime (Kessler, Ruscio, Shear, & Wittchen, 2010), thoroughly understanding anxiety symptom development prior to the typical age of onset is essential. Understanding how anxiety symptoms vary across developmental stages in individuals with ASD can offer a basis for comparing the trajectories of anxiety development across typically-developing and ASD populations.

The collection of large-scale prevalence data in ASD across different ages has been limited, and thus no reports of the estimated onset for anxiety disorders in ASD are yet available. However, developmental changes in anxiety in individuals with ASD have been studied through five studies (Bitsika & Sharpley, 2015; Davis et al., 2011; Gotham, Brunwasser & Lord, 2015; Teh, Mei-En, Chan, Ke Jia Tan & Magiati, 2017; Vasa et al., 2013). All four studies indicated that the levels of anxiety symptoms were lowest in early childhood and showed variation across the lifespan. When considered alongside the risk for developing an anxiety disorder in middle childhood and adolescence for the general population, these studies suggest that middle childhood is a transitional period wherein individual changes in anxiety merit closer study in ASD.

As with developmental changes in anxiety in ASD, little is known about what predicts the development of anxiety in ASD. In non-ASD children, a family history of anxiety disorders, a behaviourally inhibited temperament, and various parenting behaviours in early childhood are risk factors for the development of anxiety disorders (Creswell, Murray, Stacey, & Cooper, 2011; Degnan, Almas, & Fox, 2010; Gregory & Eley, 2011). In children with ASD, this research has been extended through retrospective and cross-sectional studies. First-degree relatives of individuals with ASD are more likely to have anxiety disorders and depression (Bolton, Pickles, Murphy, & Rutter, 1998; Kuusikko-Gauffin et al.,
2012; Micali, Chakrabarti, & Fombonne, 2004). However, whether parent anxiety is positively associated with child anxiety in ASD is unclear. Cross-sectional studies regarding the association between parent anxiety and adolescent anxiety have reported mixed findings (Conner, Maddox, & White, 2012; Mazefsky, Conner, & Oswald, 2010).

A challenge to further understanding anxiety in ASD is the limited knowledge around appropriate anxiety measurement in the ASD population. Many studies to date have relied on previously existing anxiety instruments developed for assessing anxiety in non-ASD populations (Vasa et al., 2014). However, the psychometric properties of most existing anxiety rating scales were originally established with children who are typically-developing or have anxiety disorders, and cannot be assumed to measure the same constructs in the ASD population (Grondhuis & Aman, 2012). Several factors specific to ASD, such as communication difficulties and alexithymia, may impact the reliability and validity of such measurement tools (Grondhuis & Aman, 2012). Evaluating the appropriateness of a measure for the population of focus is thus an essential preliminary step to answering research questions regarding anxiety in children and youth with ASD (Grondhuis & Aman, 2012; Kerns & Kendall, 2012).

**Research Objectives**

The aim of this dissertation was to add to the understanding of anxiety disorders in children with ASD through an examination of the nature, origins, and changes in anxiety symptoms during middle childhood in ASD. To this end, the specific objectives of this dissertation were to: (1) examine the psychometric properties and nature of ASD through examination of an existing rating scale; (2) examine changes in anxiety symptoms during middle childhood in ASD; and (3) examine whether sex, language ability, as well as parental
psychopathology and parent-rated anxiety in early childhood are predictive of later parent-rated anxiety symptoms in ASD.

**Research Studies, Questions, and Hypotheses**

**Study 1.** In Study 1 the factor structure, convergent and discriminant validity of a parent-report anxiety measure, the Spence Children’s Anxiety Scale – Parent Report (SCAS-P; Nauta et al., 2004) were evaluated in a sample of children ages 7.5 to 11 with ASD ranging in cognitive functioning.

**Research question 1.** Does the original factor structure of the SCAS-P show fit in a sample of 7 to 11 year old children with ASD ranging in IQ?

**Research question 2.** If not, what are the areas of inadequate fit in the SCAS-P original structure and do its individual subscales capture anxiety experiences within this sample?

**Research question 3.** Do individual SCAS-P subscales show structural validity, internal consistency and convergent and discriminant validity with a previously validated scale of emotional and behavioural functioning in children with ASD?

**Rationale and hypotheses.** The SCAS-P is a free, publically available anxiety rating scale based on a complementary child-report version (Spence, 1999). It is commonly used for clinical, community screening, and research purposes (Nauta et al., 2004), and has been used across a number of studies of children with ASD (Grondhuis & Aman, 2012; Wigham & McConachie, 2014). However, validation of the SCAS-P for use in the ASD population is in its early stages. The SCAS-P’s relation to other variables has been reported in an ASD sample (Zainal et al., 2014), and its original factor structure was not a good fit for a large sample of children with ASD (Magiati et al., 2016). The aim of Study 1 was to expand on
the psychometric evaluation of the SCAS-P in this population, by further examining its internal structure in children with ASD between ages 7.5 to 11 ranging in cognitive functioning, and evaluating its convergent and discriminant validity with a general measure of child psychopathology.

The validation study was informed by previous SCAS-P research. Evidence for the structural validity of the SCAS-P originally came from community and clinical samples in the general population from Holland and Australia (Nauta et al., 2004). Follow-up structural validation of the SCAS-P has been conducted on community samples in Turkey (Orbay & Ayvasik, 2006), Hong Kong (Li, Lau, & Au, 2011), and Japan (Ishikawa et al., 2013), as well as in both community and clinical samples in Denmark (Arendt, Hougaard, & Thastum, 2014) and the United States (Whiteside & Brown, 2008). These studies have supported a six-correlated factor model, reflective of the Diagnostic and Statistical Manual of Mental Disorders (4th ed.; DSM-IV; American Psychiatric Association [APA], 2000) classification of childhood anxiety disorders. Magiati et al. (2016) found the six-correlated factor structure to be a poor fit in a sample of 870 children with ASD. It was thus hypothesized that the overall six-correlated factor structure would not show a good fit in the current sample.

Similarly, it was hypothesized that individual subscales within the SCAS-P reflecting anxiety disorders that overlap with symptoms of ASD (i.e. OCD; Social Anxiety), would not show evidence of fit, based on past structural validation studies in this population (Hallett, Lecavalier, et al., 2013; Magiati et al., 2016; Stern et al., 2014; White et al., 2014). However, it was hypothesized that the remaining subscales (i.e., Generalized Anxiety, Separation Anxiety, Specific Phobia, Panic and Agoraphobia) would show good fit.
Study 2. Based on the structural support for Generalized, Separation Anxiety, Agoraphobia and Panic symptom factors in Study 1, Study 2 used these subscales to examine anxiety at snapshots in time (i.e., at age 7.5, 8.5, 9.5 and 10.5), and also as changing over time (i.e., over the period of middle childhood from ages 7 to 11). Additional models examined whether parent-rated anxiety in early childhood, parent internalizing symptoms, language ability, and sex predict, or explain variability in change in anxiety over this time.

Research question 1. What is the level of parent-rated Generalized, Separation Anxiety, Panic and Agoraphobia symptoms experienced at each of ages 7.5-8, 8.5-9, 9.5-10, and 10.5-11? What proportion of children showed “Elevated” levels of parent-rated Generalized and Separation Anxiety based on SCAS-P norms?

Research question 2. What is the trajectory of parent-rated Generalized, Separation Anxiety, Panic and Agoraphobia symptoms over middle childhood (age 7 to 11 years)?

Rationale and hypotheses. The estimated rates of elevated anxiety in ASD vary widely across studies, ranging from 11%-84% (White, Oswald, Ollendick, & Scahill, 2009). When considered altogether, previous research suggests that approximately 40% of children with ASD up to age 18 meet criteria for an anxiety disorder based on exceeding clinically significant levels on rating scales, or based on clinical interview (van Steensel, Bögels, & Perrin, 2011). Moreover, clinically significant symptoms of Generalized Anxiety, Separation Anxiety, Panic Disorder and Agoraphobia across studies were estimated at 15.4%, 9%, 1.8% and 16.6% respectively (van Steensel et al., 2011). Although several studies have provided broad prevalence estimates of anxiety specific to narrow age ranges (e.g., Niditch, Varela, Kamps, & Hill, 2012: ages 2-3; 4-5; 6-9; Weisbrot, Gadow, DeVincent, & Pomeroy, 2005: ages 3-5; 6-12), most studies to date have reported single rates for wide age ranges (e.g., ages
5-17: Leyfer et al., 2006; Sukhodolsky et al., 2008). In Study 2, anxiety at four single, concentrated developmental periods (i.e., age 7.5, 8.5, 9.5 and 10.5) was measured, allowing for a finer-grained understanding of its occurrence in middle childhood.

The expression of both disordered and adaptive anxiety changes with age (Bongers, Koot, van der Ende, & Verhulst, 2003; Kessler, Petukhova, Sampson, Zaslavsky, & Wittchen, 2012; van Oort, Greaves-Lord, Verhulst, Ormel, & Huizink, 2009). Moreover, typically developing children demonstrate distinct developmental risk periods for anxiety disorders (Kessler et al., 2005). Thus, it was expected that trajectories of anxiety symptoms in ASD would show change across middle childhood. Thus, trajectories of parent-rated Generalized Anxiety, Separation Anxiety, Panic Disorder and Agoraphobia symptoms across ages 7 to 11 were examined by research question 2. The majority of previous research has relied on cross-sectional samples to examine changes in various aspects of anxiety across time. Bitsika and Sharpley (2015) focused on changes in each two-year span from ages 6 to 18, while Vasa et al. (2013) and Davis et al. (2011) compared broader age spans from the early years of life (ages 1 to 6) through to late adulthood (up to 65). These studies all found lower levels of anxiety at younger ages, though reported different patterns of change from childhood through adolescence. One study of children ages 5-17 who were followed up 10-19 months later found no change in overall mean anxiety scores, though noted that 20% of the sample showed a change in severity of anxiety levels across the two time points (Teh et al., 2017). Research question 2 added to this literature using longitudinal data to examine the overall change in anxiety over a short developmental period in middle childhood, and examined whether the amount of change varies across children. Based on the above studies, it was hypothesized that levels of parent-rated Generalized Anxiety, Panic Disorder
symptoms and Agoraphobia would increase in the sample from ages 7 to 11. Based on decreasing incidence of Separation Anxiety in typically developing children across this time period (Kessler et al., 2005), it was hypothesized that parent-rated Separation Anxiety would decrease. Furthermore, given that shifts in the severity of anxiety symptoms have been reported across this period (Bitsika & Sharpley, 2015; Vasa et al., 2013), the amount of change in anxiety with age was expected to vary across children.

**Research question 3.** Do A) language ability B) sex C) parent internalizing symptoms and D) parent-rated child anxiety in early childhood account for a significant amount of variance in the tendency towards parent-rated Generalized Anxiety, Separation Anxiety, Panic and Agoraphobia levels between ages 7-11?

**Rationale and hypothesis.** Numerous studies have reported a concurrent association between high verbal expression abilities, IQ or adaptive abilities, and severity of anxiety. However, findings have been mixed, with some studies reporting that higher-functioning children experience more anxiety (Hallett, Lecavalier, et al., 2013; Lecavalier, 2006; Mazurek & Kanne, 2010; Sukhodolsky et al., 2008; Weisbrot et al., 2005) and others finding no association (Simonoff et al., 2008; Strang et al., 2011). A positive relationship between verbal ability and anxiety is expected in part because verbally expressed distress, anxiety, or worry, or reports of physical symptoms are one way for others to recognize anxiety as a problem (Wood & Gadow, 2010). Moreover, children typically begin to use emotional vocabulary (e.g., “I am scared; I am angry”) after they develop phrase speech (Sparrow, Cicchetti & Balla, 2005), suggesting that there is likely to be a graduated correlation between a child’s language ability and parents’ observations of expressed anxiety. Thus, research question 3 examined whether language abilities in middle childhood predict anxiety levels,
and/or account for a significant proportion of the variance in change in parent-rated anxiety from ages 7-11. It is hypothesized that language ability will be significantly predictive of parent-rated anxiety, and account for a significant amount of variance in change over time.

A lack of longitudinal research on anxiety in ASD has limited research on predictive factors. However, evidence from otherwise typically-developing children suggests that being female, as well as a number of familial or behavioural factors in early childhood are risk factors for the later development of anxiety (Degnan et al., 2010). There is indirect evidence that a family history of internalizing disorders is also related to anxiety in ASD (Bolton et al., 1998; Kuusikko-Gauffin et al., 2012; Micali et al., 2004). However, a direct examination of how parental psychopathology and anxiety symptoms in early childhood predict the development of anxiety disorders in children with ASD is missing from the literature. Though several studies have reported no sex differences in anxiety, most samples have included small numbers of females (Gotham et al., 2015; May, Cornish, & Rinehart, 2014; Park et al., 2012; Simonoff, Pickles, Charman, Chandler, Loucas & Baird, 2008; Solomon, Miller, Taylor, Hinshaw & Carter, 2011). It was hypothesized that no sex differences would be apparent in this sample, but that both parent-rated anxiety in early childhood, and parent internalizing symptoms at the time of the child’s diagnosis would be predictive of anxiety in middle childhood.

Summary

Anxiety disorders are highly prevalent and impairing to children with ASD (van Steensel et al., 2011; White et al., 2009). Research in this area relies on psychometrically sound anxiety measurement tools. However, existing anxiety measures may not be appropriate for use with ASD populations. Moreover, while there is significant research
around early anxiety development in the general population (Kessler et al., 2010; Rapee, Schniering, & Hudson, 2009), such research is limited in children with ASD. Research that closely examines changes in anxiety symptoms during concentrated early developmental periods is needed to suggest an appropriate developmental period for preventative interventions. Further research into early predictors could inform early identification of those at risk for developing anxiety disorders. This dissertation was composed of two studies. The first study looked at evidence for the validity and internal consistency of the SCAS-P when used with this population. The second study closely examined parent-rated anxiety symptoms as they occur from ages 7 to 11, and evaluated whether language ability, sex, parent internalizing psychopathology and early parent-rated anxiety symptoms were predictive of a tendency towards anxiety in middle childhood.
Chapter 2: Literature Review

This chapter presents a brief overview of ASD, along with an overview of common comorbid medical disorders and psychopathologies and their incidence in individuals with ASD. Next, a discussion of normative anxiety and clinically significant anxiety disorders as they occur in the general population is presented. A section that introduces the literature on anxiety disorders in ASD follows, including their prevalence, conceptualization, presentation and impact. Challenges to anxiety assessment unique to the ASD population, and a review of structural validation studies of existing anxiety rating scales with ASD samples are presented in the introduction to Study 1 (Chapter 3). A review of the literature on predictors of anxiety in children with ASD, as well as of developmental changes in anxiety in ASD precedes Study 2 (Chapter 4).

Autism Spectrum Disorder

Autism spectrum disorder is characterized by a qualitative impairment in social communication and repetitive behaviours or circumscribed interests (APA, 2013). Symptoms of ASD are heterogeneous in presentation ranging from mild to severe levels of social impairment, repetitive behaviours, language, cognitive ability, and aberrant behaviour (Volkmar et al., 2014). There is considerable evidence that ASD is a neurodevelopmental condition with a significant genetic component (Geschwind, 2011; Kim & Leventhal, 2015). However, no biological test or biomarker for ASD has yet been identified (Kim & Leventhal, 2015). ASD is significantly more prevalent in males than females, with a striking and unexplained 4:1 ratio (Baio, 2014; Baron-Cohen et al., 2011). Data from 43 prevalence surveys across the world using Diagnostic and Statistical Manual of Mental Disorders (DSM-IV-TR; 4th ed., text rev; APA, 2000) criteria for pervasive developmental disorders indicated prevalence estimates ranging from 60-70 individuals per 10,000 (Fombonne, 2009).
Moreover, ASD is estimated to have increased twenty to thirtyfold since the 1960’s or 70s worldwide (Baio, 2014; Elsabbagh et al., 2012) although some uncertainty remains around the exact prevalence and rising rate (Fombonne, 2009). This dramatic surge has led to regular population-level surveying by the Centre for Disease Control and Prevention Autism and Developmental Disabilities Monitoring Network across the United States since 2002 (Baio, 2014). Their most recent estimate from 2010 indicated that one 8-year-old child for every 68 children in the United States is affected (Baio, 2014).

**Comorbidity.** Comorbidity, or the co-occurrence of two or more conditions, is common across all childhood psychiatric disorders and is particularly common in ASD (Costello, Mustillo, Erkanli, Keeler, & Angold, 2003). The assessment of comorbid conditions in ASD is inherently challenging due to its core communication deficits and frequently associated intellectual impairment (Matson & Nebel-Schwalm, 2007; Simonoff et al., 2008; Volkmar et al., 2014). Furthermore, efforts to determine the prevalence of comorbid conditions have been hampered by methodological issues, and resulting estimated rates often span a wide range. With these caveats in mind, estimates of the prevalence of comorbid medical and psychiatric conditions in ASD are summarized below.

With regard to medical comorbidity, sleep disorders, gastrointestinal disorders and seizure disorders are highly prevalent in ASD, and estimated as occurring in 40%-80%, 9%-91%, and 10%-40% of children, respectively (Bauman, 2010; Johnson, Giannoti, & Cortesi, 2009; Mannion & Leader, 2013). Intellectual disability is also common in individuals with ASD, with the most recent estimates suggesting that 23% to 31% of children diagnosed with ASD function in the intellectually impaired range (i.e., IQ < 70; Baio, 2014; Idring et al., 2015). Externalizing and internalizing psychopathology is also common in this population.
Estimates of Attention Deficit/Hyperactivity Disorder (ADHD) range widely from 5%-16% (Hanson et al., 2012; Keen & Ward, 2004) to 50%-78% (Gadow, DeVincent, Pomeroy, & Azizian, 2004; Gadow, Devincent, Pomeroy, & Azizian, 2005; Lee & Ousley, 2006; Sinzig, Walter, & Doepfner, 2009; Yoshida & Uchiyama, 2004). The rate of Oppositional defiant disorder has been estimated at 7%-28% of children (Leyfer et al., 2006; Simonoff et al., 2008). The rate of Depression is estimated at 2%-30% of cases (Ghaziuddin & Mikhail, 1998; Leyfer et al., 2006; Simonoff et al., 2008). Bipolar disorder has also been reported, but never in more than one individual within a sample (Leyfer et al., 2006) or in a case study (Gillberg, 1985). Approximately 40% of children and youth with ASD are estimated to meet criteria for one or more anxiety disorders (van Steensel et al., 2011). Anxiety is one of the most common psychopathologies in ASD and is the focus of the remaining literature review.

**Anxiety**

Anxiety is a fearful response that includes cognitive, physiological, and behavioural components (Lang, 1968). Cognitively, anxiety is characterized by worried thoughts and apprehension: these may include negative automatic thoughts or cognitive errors, including catastrophic predictions, all-or-nothing thinking, making assumptions about other people’s internal thoughts or feelings, anticipating failure to cope, and exaggerating or minimizing the relative importance of positive or negative experiences (Pêgo, Sousa, Almeida, & Sousa, 2010). Physiologically, anxiety has somatic effects, including increased heart rate, dizziness, light-headedness, muscle tension and sweating (Pêgo et al., 2010). Behaviourally, anxiety is often expressed through avoidance; nervous behaviours such as trembling or truncated speech; and safety behaviours that facilitate coping, such as seeking reassurance or hyper-vigilance (Risbrough, 2010). Importantly, early research demonstrated that cognitive,
behavioural and physiological responses do not reflect a unitary experience, but rather vary in intensity across these three domains (Lang, 1968). For example, a typically developing individual’s autonomic response to a feared stimulus may differ substantially from the cognitive experience that they describe through verbal report (Lang, 1968). The incongruity between an individual’s physical and cognitive experiences of anxiety, along with the frequent reliance on rating scales and interviews in clinical and research assessment, has led some researchers to suggest that rating scales may inadvertently bias assessment towards the cognitive features of anxiety (Risbrough, 2010).

Experiences of anxiety range on a continuum of severity and are often adaptive in that they offer protection from a perceived threat (Pêgo et al., 2010). Indeed, elevated anxiety is normative at some stages of development. For example, preschoolers commonly show fears of physical injury from dogs, spiders, or thunderstorms, as well as separation anxiety or fear of sleeping alone (Spence, Rapee, McDonald, & Ingram, 2001). Anxiety becomes maladaptive when it is excessive in severity, persists over time, and has a significant impact on functioning (Pêgo et al., 2010; Rapee et al., 2009). Non-adaptive anxiety, physiological arousal to triggering stimuli, and avoidance are shared features of all anxiety disorders (APA, 2013). Anxiety disorders include different symptom constellations distinguishable based on the focus of anxiety, perceived feared outcomes and behavioural differences (Rapee et al., 2009).

**Prevalence.** The estimated prevalence of anxiety disorders in children across large-scale epidemiological studies is 3.8% (Waddell, Shepherd, & Schwartz, 2014). Children with some chronic medical conditions are at a much higher risk of developing comorbid anxiety disorders (Pinquart & Shen, 2011). The strongest risk is associated with chronic
fatigue, migraines, sensory impairment disorders and epilepsy, conditions in which the emergence of symptoms and the overall course of illness are largely outside of the individual’s control (Pinquart & Shen, 2011).

In typically developing children, estimates for the prevalence of specific anxiety disorders range widely. Separation anxiety disorder affects 4%-5% of children ages 6-8 (Masi, Mucci, & Millepiedi, 2001). Social anxiety disorder affects approximately 9.1% of children and youth (Merikangas, He, Burstein, et al., 2010), and GAD is estimated at 2.2% (Merikangas, He, Burstein, et al., 2010). Other anxiety disorders are less common, with panic disorder estimated at a prevalence of 0.7% (Merikangas, He, Brody, et al., 2010), agoraphobia at 2.4% (Merikangas, He, Burstein, et al., 2010), and OCD at 0.25-0.4% (Heyman et al., 2003; Waddell et al., 2014).

**Lifetime risk.** A fuller picture of anxiety disorder prevalence is given by estimates of the proportion of individuals who will develop an anxiety disorder at some point in their lives. Recent U.S. epidemiological data have reported the lifetime morbid risk (LMR), or the estimated probability that one will develop an anxiety disorder during their lifetime (Kessler et al., 2012). These data suggest that the most common anxiety disorders are specific phobia (18.4% LMR), social anxiety disorder (13.0% LMR), GAD (9.0% LMR), separation anxiety disorder (8.7% LMR), and panic disorder (6.8% LMR; Kessler et al., 2012). Agoraphobia and OCD are the least common (2.7%-3.7% LMR: Kessler et al., 2012). Furthermore, the prevalence of anxiety disorders is on the rise and is increasing more dramatically than for any other class of mental health disorders (World Health Organization, 2000). Within the anxiety disorders, historical increases in prevalence have been more pronounced for social anxiety disorder and GAD than other anxiety disorders (Kessler & Greenberg, 2002).
**Age and developmental changes.** Anxiety disorders have an earlier age of onset than other classes of mental health disorders, including mood and substance abuse disorders (Kessler et al., 2005). International surveys estimate the median age of onset for anxiety disorders to be between 11 (Kessler et al., 2005) and 15 years of age (World Health Organization, 2000). Moreover, the distribution of age of onset is relatively concentrated for anxiety disorders: 50% of cases become apparent between ages 6-21 (Kessler et al., 2005). This pattern contrasts to most chronic physical health conditions for which the risk of occurrence increases with age, and for which the range of age of onset has an upper limit in late adulthood or old age (Murray & Lopez, 1996). Taken together, these data suggest that anxiety disorders pose the highest risk in childhood and adolescence, with the odds of developing an anxiety disorder substantially lower later on in life (Kessler et al., 2005).

Within the anxiety disorders, the age of onset distributions vary. Specific phobias and separation anxiety disorder tend to begin in early childhood, with a median onset at age 7 (Kessler et al., 2005), with particularly concentrated ranges of onset (ages 5-12; ages 6-11; respectively). Other anxiety disorders become more problematic during puberty and into adolescence: social anxiety disorder tends to begin in early adolescence (median onset at age 13), with a similarly narrowly distributed range (ages 8-15). Panic disorder, OCD, and GAD have later ages of onset (median age 19-31) and wider distributions in ages of onset (ages 14-30; Kessler et al., 2005).

**Comorbidity.** Anxiety disorders are highly comorbid with one another, with up to 75% of anxious children presenting with more than one anxiety disorder (Rapee, 2012). The risk of increasing psychopathology (i.e., added comorbidity) within the anxiety disorders depends on the principal diagnosis, ranging from most to least as follows: GAD, panic
disorder with agoraphobia, social anxiety disorder, and specific phobia (Brown, Campbell, Lehman, Grisham, & Mancill, 2001). That is, a diagnosis of specific phobia is the least likely to present alongside other severe symptoms of anxiety, while GAD holds the highest risk. Moreover, anxiety disorders are also commonly comorbid with other psychopathologies (Costello et al., 2005; Kessler et al., 2010). Importantly, anxiety disorders are often the first to onset in individuals with multiple comorbid psychopathologies (Kessler et al., 2010). This sequence suggests that the presence of an anxiety disorder is a risk factor for the development of secondary disorders, and highlights the potential preventative effect of targeting childhood and adolescent anxiety (Wittchen, Kessler, & Pfister, 2000).

**Persistence.** Although treatment studies indicate that many childhood anxiety disorders can remit within three to four years (Last, Perrin, Hersen, & Kazdin, 1996; Walkup et al., 2008), epidemiological surveys suggest anxiety disorders are the most persistent group of psychopathologies (Kessler et al., 2010; 2012). As a group, anxiety disorders persist through the lifetime, showing only a very modest decline into old age (Kessler et al., 2012). The pattern of persistence differs, however, with age and across the anxiety disorders (Kessler et al., 2012). For example, specific phobia shows a persistent course into old age; GAD tends to show a chronic, but episodic course; and only about 20% of child-adolescent onset separation anxiety disorders persist beyond adolescence (Kessler et al., 2012).

**Predictors of anxiety.**

**Sex.** Most anxiety disorders present more commonly in females, and this pattern persists into adulthood (McLean & Anderson, 2009). Girls are twice as likely as boys to develop an anxiety disorder by age 6 (Lewinsohn, Gotlib, Lewinsohn, Seeley, & Allen, 1998) and into adolescence (Kessler et al., 2012). Within the anxiety disorders, girls are more
likely to experience separation anxiety disorder and agoraphobia (Costello et al., 2003; Kessler et al., 2012; Merikangas, He, Burstein, et al., 2010). Some reports suggest that GAD follows this same pattern (Merikangas, He, Burstein, et al., 2010), while other surveys indicate that GAD only becomes more prevalent in females in adulthood (Kessler et al., 2012). Social anxiety disorder is more prevalent in adolescent girls (Merikangas, He, Burstein, et al., 2010), though the disparity subsides in adulthood (Bekker & van Mens-Verhulst, 2007; McLean, Asnaani, Litz, & Hofmann, 2011). The sex difference is reversed in childhood OCD, which is more common in boys, though its incidence evens out in adolescence and adulthood (Bekker & van Mens-Verhulst, 2007; Castle, Deale, & Marks, 1995).

**Family factors.** In typically-developing children, genetic, temperamental and environmental factors are early risk factors for anxiety disorders in childhood (Degnan et al., 2010; Gregory & Eley, 2007; van der Bruggen, Stams, & Bögels, 2008). Genetics account for about 30% of the variance in childhood anxiety disorders (Gregory & Eley, 2007). The degree of heritability is higher for girls than boys (Eaves et al., 1997; Feigon, Waldman, Levy, & Hay, 2001) and also depends on the age of onset (Gregory & Eley, 2011). It is further estimated that shared environments (e.g., socioeconomic status) account for an additional 20% of the variance and unique environmental influences account for the remaining 50% (Gregory & Eley, 2007). Whether the anxiety disorders share a general heritable component or whether familial transmission is specific within the anxiety disorders is unclear due to contrasting evidence from population and twin-studies (Rapee, 2011; Gregory & Eley, 2011).

In addition to heritability, anxiety symptoms in middle childhood are also predicted
by a behaviourally inhibited temperament in early childhood (Degnan et al., 2010). Behavioural inhibition is expressed as a reticent temperament and, depending on the child’s developmental stage, can include emotional negativity, withdrawn or cautious responding to novelty and extreme social reticence (Degnan et al., 2010). Although a behaviourally inhibited temperament occurs in 15%-20% of children (Fox, Henderson, Marshall, Nichols, & Ghera, 2005), only some of these children will go on to develop anxiety disorders (Degnan et al., 2010). Nonetheless, a persistently inhibited temperament can put children at risk for specific phobias (Hirshfeld et al., 1992) and social anxiety disorder (Chronis-Tuscano et al., 2009; Hirshfeld-Becker & Biederman, 2007; Muris, van Brakel, Arntz, & Schouten, 2011).

Many parenting behaviours and family dynamics have also been posited as contributing to the development of childhood anxiety (Creswell et al., 2011; Drake & Ginsburg, 2012). Although evidence for the majority of hypothesized influential factors has shown inconsistent or very small effects, two risk factors have consistently emerged from this literature (Drake & Ginsburg, 2012). The first of these is an insecure attachment style in early childhood (Colonnesi et al., 2011; Warren, Huston, Egeland, & Sroufe, 1997). An insecure attachment can result from insufficient or inconsistent parental attunement and sensitivity to the cues of the child or infant (Ainsworth, Blehar, Waters, & Wall, 1978). Colonnesi et al.’s (2011) meta-analysis of 46 studies using both longitudinal and cross-sectional analyses reported a moderate and significant relationship between insecure attachment and anxiety in childhood.

The second risk factor is parental overcontrol, or the pressure parents place on their child to act, think, or feel in certain ways. Parental overcontrol has been shown to have a medium effect size on childhood anxiety disorders (van der Bruggen et al., 2008).
Overcontrol can be exerted through overprotective parenting that prevents a child from developing their own coping skills in new situations (Barlow, 2002), or by influencing a child’s perception of threat (Rapee, 2001). However, like most environmental influences (including parent-child attachment), it is important to consider that parental over control is bidirectional: it may be both triggered and maintained by the child’s behaviour (Rapee, 2001). Thus, it is not surprising that the relationship was moderated by various factors, with the strongest effects in studies with an overrepresentation of girls, school-age children, and families with a high socioeconomic status (van der Bruggen et al., 2008).

**Anxiety in Children and Youth with ASD**

**Prevalence.** In the first published review of the literature on the prevalence of anxiety disorders in children with ASD, 11% to 84% of children with ASD were reported to experience impairing levels of anxiety across 11 identified studies (White et al., 2009). An updated review of 24 studies found a similar range in estimates and suggested the source of participants had the biggest influence on estimates, with higher rates reported in clinical samples or studies that advertised their focus on anxiety (Kerns & Kendall, 2012).

Furthermore, a meta-analysis of 31 studies estimated that 39.6% of children and youth with ASD meet criteria for at least one anxiety disorder (van Steensel et al., 2011). Van Steensel et al. (2011) also aggregated estimates for different anxiety disorders in children and youth with ASD. This report suggested that panic disorder was the least prevalent, occurring in 1.8% of children and youth; separation anxiety disorder occurred in 9%, GAD occurred in 15.4%, social anxiety disorder and agoraphobia occurred in 16.6%, OCD occurred in 17.4%, and specific phobias occurred in 29.8% (van Steensel et al., 2011). Like the overall
prevalence rate, van Steensel et al.’s reported rates are also strikingly higher than the prevalence in typically-developing children and youth described previously.

**Impact.** Some parents of children with ASD have reported that the impact of their child’s anxiety is more deleterious than that of core ASD symptoms (Ozsivadjian & Knott, 2011; Ozsivadjian et al., 2012). Consistently, anxiety in ASD is associated with a decreased quality of life (van Steensel, Bögels, & Dirksen, 2012). Comorbid anxiety has been associated with increased repetitive speech, repetitive behaviours and focus on circumscribed interests (Magiati et al., 2015), as well as increased sleep and gastrointestinal problems (Williams, Leader, Mannion, & Chen, 2015). Social functioning is also negatively impacted, particularly by social anxiety, which creates social fear and avoidance and in turn prevents the practice of social skills (Chang, Quan, & Wood, 2012; White et al., 2010).

**Conceptualization.** Speculation about the association between anxiety and ASD began with the earliest observations of autism by Kanner (1943), whose case studies describe several children with extreme fears of various objects and situations. Empirical studies have since demonstrated that the association between autistic symptomatology and anxiety is similarly applicable outside of clinical ASD. A positive relationship between ASD symptoms and anxiety symptoms has been shown in children and youth with mood and anxiety disorders, but without ASD (Pine, Guyer, Goldwin, Towbin, & Leibenluft, 2008; Settipani, Puleo, Conner, & Kendall, 2012).

The conceptualization of anxiety disorders as a clinically impairing group of disorders that are distinct from, but sometimes adjunct to, ASD is quite recent. Anxiety was initially understood to simply be part of the autistic profile, even though anxiety in severe presentations was considered to merit targeted treatment (Lainhart, 1999). The historical
change in understanding has been reflected in the *DSM*’s suggested criteria: until the most recent revision (APA, 2013), the diagnostic criteria suggested that ASD should not be diagnosed concurrently with GAD, separation anxiety, or social phobia (APA, 2000). The ambiguity regarding whether anxiety in ASD is distinguishable from the core symptoms of ASD has been, in part, due to significant overlap in the symptomatology of the two disorders (Kerns & Kendall, 2012; Skokauskas & Gallagher, 2010; Wood & Gadow, 2010).

Symptoms including obsessive thinking patterns; ritualistic, repetitive, or compulsive behaviour; excessive questioning; social avoidance; and reduced emotional reciprocity can potentially be attributed to either anxiety disorders or ASD (Kerns & Kendall, 2012; Wood & Gadow, 2010).

**Presentation.** The nascent conceptualization of anxiety in ASD is also reflected in the variety in descriptions and approaches to measuring anxiety used across the literature (White et al., 2009). Capturing the presentation of anxiety in ASD requires an understanding of the antecedents, maintaining factors, and expressions of anxiety that may be unique or may be shared with anxiety disorders in otherwise typically developing individuals (Rudy, Lewin, & Storch, 2013). Only one study to date has attempted to shed light on this issue in children through an understanding of parental perspectives (Ozsivadjian et al., 2012). Through focus groups with 17 parents of children with ASD, parents identified the two most common triggers to anxiety as social situations and change or interruption to routines (Ozsivadjian et al., 2012). Examples included small changes to routine such as being stuck in traffic, as well as non-routine events that others would typically consider enjoyable such as holidays or birthdays (Ozsivadjian et al., 2012). Situational social triggers included common worries such as being the focus of attention, fear of embarrassment or ridicule, and fears
related to self-awareness of social difficulties (Ozsivadjian et al., 2012). Additional triggers included sensory stimuli, such as certain noises or smells, and overwhelming expectations around performance or organization. Specific fear-provoking stimuli have also been reported to trigger anxiety in children with ASD, including commonly feared things (e.g., spiders) as well as some idiosyncratic and situations (e.g., walking over a bridge, crowded vehicles; Evans, Canavera, Kleinpeter, Maccubbin, & Taga, 2005; Ozsivadjian et al., 2012). Some overlap between typically-developing populations and those with ASD has been reported in the cognitive factors that serve to maintain anxiety: negative automatic thinking, including catastrophizing; fortune-telling; and all-or-nothing thinking have been identified, though at lower rates, in adolescents with ASD (Farrugia & Hudson, 2006; Ozsivadjian et al., 2012).

A study that compared the anxiety-provoking and worrisome thoughts of adults with Asperger Syndrome to typically-developing adults using real-time recording further characterizes the cognitive features of anxiety in ASD (Hare, Wood, Wastell, & Skirrow, 2015). In the Asperger Syndrome group, ruminating on thoughts for over 10 minutes and having a train of thought be interrupted were associated with feelings of anxiety (Hare et al., 2015). Interestingly, for adults with Asperger Syndrome, no relation was found between the topic of their thoughts and their level of anxiety, though worrisome thoughts were more likely to be internally focused. Of reported anxious thoughts, 78% were about general events (e.g., worrying about the bus arriving late), 15.7% were related to fear of negative evaluations by others, and 6% were health-related (Hare et al., 2015). Similar to typically-developing adults, 52% of the worrisome thoughts for adults with Asperger Syndrome occurred when the individuals were alone (Hare et al., 2015).
The expression of anxiety in children with ASD differs from that of typically developing children in a number of ways (Ozsivadjian et al., 2012). First, anxiety in children with ASD is commonly more evident through behaviour than through children’s reports of worries, fears, or somatic complaints. Parents describe that anxiety is evidenced in challenging behaviours such as verbal or physical aggression; avoidance, withdrawal, and/or escape from the anxiety-provoking situation; increased activity levels; sensory seeking behaviours; and increased repetitive or stereotyped behaviours (Ozsivadjian et al., 2012). Parents also described that their children’s anxiety was also apparent through somatic signs, with disruptions to sleep most commonly reported (Ozsivadjian et al., 2012). Moreover, youth with ASD were reported to show an unusual delay between an anxiety-provoking situation and the expression of anxiety, making pre-emptive anticipation of anxiety challenging for outside observers (Ozsivadjian et al., 2012).

**Summary**

This literature review provided a foundation for the two research studies conducted for this dissertation. The chapter included discussions of ASD, anxiety disorders, and research about their comorbid presentation. There is substantial evidence that ASD and anxiety disorders are commonly comorbid (van Steensel et al., 2011; White et al., 2009). However, their overlapping features have complicated how anxiety has been conceptualized in relation to ASD and differential assessment (Kerns & Kendall, 2012).
Chapter 3: Study 1

Comorbid anxiety disorders occur in approximately 38% of children and youth with autism spectrum disorder (ASD; van Steensel, Bögels, & Perrin, 2011). Elevated anxiety symptoms cause impairment beyond that caused by the core symptoms of ASD (Ozsivadjian, Knott, & Magiati, 2012). Anxiety measures that are specifically designed to accommodate ASD symptomatology (e.g., intolerance of uncertainty, sensory fears) have been developed only recently (e.g., Rodgers et al., 2016). Hence, general anxiety measures originally developed for individuals without ASD, including the Spence Children’s Anxiety Scale – Parent report (SCAS-P; Spence, 1999), are still widely used to study the phenomenon of anxiety in those with ASD (e.g., Teh et al., 2017; Ooi et al., 2016).

However, anxiety assessment in ASD is complicated. One challenge is the overlap in symptomatology between anxiety disorders and ASD (Wood & Gadow, 2010). For example, limited eye contact, social avoidance, and a preference for spending time alone are observed in social anxiety disorder as well as in ASD (Kerns et al., 2016; Wood & Gadow, 2010). Similarly, the obsessive thoughts and compulsive behaviour observed in obsessive-compulsive disorder (OCD) – classified as an anxiety disorder until the 2013 publication of the Diagnostic and Statistical Manual of Mental Disorders (5th ed.; DSM–5; American Psychiatric Association, 2013) – can be confused with the repetitive behaviour, perseverative thoughts, and insistence on sameness thoughts observed in ASD (Wood & Gadow, 2010). Consequently, the measurement structure of existing scales may vary based on whether anxiety symptoms present in the context of an ASD diagnosis.

Additional challenges to assessment are posed by other characteristics related to ASD. Children with ASD vary greatly in their verbal abilities, with 25%-30% of children having little to no language at kindergarten entry (Anderson et al., 2007; Tager-Flusberg &
Limited language abilities complicate assessments that rely on self-report measures, particularly as most existing anxiety scales were developed for children with age-typical language comprehension (Grondhuis & Aman, 2012; Leyfer et al., 2006). In combination with limited verbal abilities, children across the autism spectrum have impaired affect recognition and differ in how they convey their thoughts, physiological experiences, or affective states to others (Grondhuis & Aman, 2012; Leyfer et al., 2006; Wood & Gadow, 2010). Given that verbally expressed distress, anxiety, worry and reports of physical symptoms are the most common ways for others to recognize anxiety, the same problem extends to parent-report measures (Grondhuis & Aman, 2012; Wood & Gadow, 2010).

Many items used on anxiety rating scales and interviews draw on expressions of anxiety that require verbal ability in typically developing children (Grondhuis & Aman, 2012; Nauta et al., 2004; van Steensel, Bögels, Magiati, & Perrin, 2014). For example, parents may be asked to rate how frequently their child reports worries or complains about their internal feelings or somatic symptoms (Grondhuis & Aman, 2012; Nauta et al., 2004; van Steensel et al., 2014). This may not be feasible for children with ASD, particularly those with communication impairments, and will influence how their anxiety symptoms are perceived and reported (Grondhuis & Aman, 2012).

A further complicating factor is the high rates of problem behaviours displayed by individuals with ASD (Hagopian & Jennett, 2014). These behaviours include aggression, tantrums, disruptive vocalizations, self-injury and destruction of property (Hanley, Iwata & McCord, 2003). The behaviours often serve as a way to escape or avoid situations (Hagopian & Jennett, 2014; Hanley et al., 2003). The complexity for assessment arises in that problem behaviour may be associated with emotional distress, or the avoidance of an...
aversive activity, and determining the cause of the behaviour is difficult. Information about the function of the behaviour, as well as observations of accompanying fearful facial expressions, emotional distress and physiological symptoms typical of anxiety (e.g., muscle tension, fast heart rate), are needed to hypothesize whether anxiety is at the root of the behaviour (Hagopian & Jennett, 2014).

In consideration of the complexity described above, the measurement properties of anxiety scales as assessed in individuals without ASD may not apply in the ASD population. Indeed, two systematic reviews that identified existing rating scales for anxiety outcomes in this population largely relied on psychometric evidence from non-ASD children and youth (Lecavalier et al., 2014; Wigham & McConachie, 2014). With this limitation, Lecavalier et al. (2014) concluded that none of the existing measures used in the ASD literature were appropriate without conditions, while Wigham and McConachie (2014) concluded that the Screen for Child Anxiety Related Emotional Disorders (SCARED; Birmaher et al., 1997), the Revised Children’s Anxiety and Depression Scale (RCADS; Chorpita, Yim, Moffitt, Umemoto & Francis, 2000), and the SCAS-P showed the most evidence of robust measurement properties. Positively, since these reviews, the RCADS was revised specifically for ASD and showed good psychometric properties in an ASD sample (Rodgers et al., 2016). The SCARED child- and parent-report versions (Stern, Gadgil, Blakeley-Smith, Reaven & Hepburn, 2014) and the SCAS-P (Zainal et al., 2014; Magiati et al., 2016) were also psychometrically evaluated in ASD samples. However, evidence of structural validity – one of five sources of validity evidence (American Educational Research Association, 2014) – is preliminary for the SCARED (Stern et al., 2014), and lacking for the SCAS-P (Magiati et al., 2016). Thus, the intention of the current study was to identify
a psychometrically sound SCAS-P structure in order to provide an alternative approach to re-examining data that were collected using this measure.

**Structural Validation Studies of Anxiety Rating Scales in ASD**

Four studies have used factor analytic approaches to seek evidence of structural validity of existing parent- and child-report anxiety scales in ASD samples. Of these four studies, only the Multidimensional Anxiety Scale for Children – Child Report (MASC-C; March, 1997) original factor structure has been found to be a good fit for children with ASD (White et al., 2014). Exploratory analysis suggested the SCARED parent- and child-report approximated four of the five factors of the original SCARED structure (Stern et al., 2014). Due to poor fit of the original structure with ASD samples, alternative structures have been proposed for two parent-rated anxiety scales based on exploratory factor analyses: the Multidimensional Anxiety Scale for Children – Parent Report (MASC-P; March, 1997; White et al., 2014), and the Child and Adolescent Symptom Inventory - Anxiety Scale (CASI-Anxiety; Hallett, Lecavalier, et al., 2013; Weisbrot et al., 2005), though both remain to be tested for fit in an independent ASD sample. Taken together, the above studies suggest that as in children without ASD, anxiety in ASD is likely multi-faceted. However, specific symptoms of anxiety as reported on existing rating scales do not clearly fall into the same delineated categories as for typically-developing kids (Hallett, Lecavalier, et al., 2013; Magiati et al., 2016; Stern et al., 2014; White et al., 2014). Thus, assuming the structural validity of existing rating scales in their original published format for the measurement of anxiety in children with ASD is premature.

Similar to the MASC-P and the CASI-Anxiety, the original structure of the SCAS-P was not found to be a good fit based on confirmatory factor analysis (CFA) with a large
sample of children (N = 870; mean age = 11.6 years, SD = 2.77 years) with ASD (Magiati et al., 2016). Despite secondary exploratory analyses (Magiati et al., 2016), no replicable alternative structure has been proposed and the areas of misfit within the SCAS-P when used with the ASD population remain to be determined. Consequently, further evaluation is needed to identify areas of misfit within the SCAS-P and an alternative structure that may have utility for psychometrically sound assessment within this population.

**Current Study**

The current study expanded on past psychometric examination of the SCAS-P in a sample of children with ASD ages 7 to 11 years. The following research questions guided this study:

1. Does the original factor structure of the SCAS-P show fit in a sample of 7 to 11 year old children with ASD ranging in IQ?
2. If not, what are the areas of inadequate fit in the SCAS-P original structure and do its individual subscales capture anxiety experiences within this sample?
3. Do individual SCAS-P subscales show structural validity, internal consistency and convergent and discriminant validity with a previously validated scale of emotional and behavioural functioning in children with ASD?

**Method**

**Participants**

Data for this study were drawn from an ongoing longitudinal study (*Pathways in ASD*) examining the developmental trajectories of children with ASD (N = 421) across five Canadian sites. Children were diagnosed with ASD between 23-59 months of age using *DSM-IV-TR* criteria (APA, 2000) by multidisciplinary clinical teams and confirmed by the Autism Diagnostic Interview-Revised (Lord, Rutter, & LeCouteur, 1994) and the Autism
Diagnostic Observation Schedule (Lord et al., 2000). Children with a diagnosis of cerebral palsy or other neuromotor disorders, identified genetic or chromosomal abnormalities, or significantly impaired vision or hearing were excluded from the study. The study was approved by the Research Ethics Board at each site.

The present study’s assessments were conducted annually beginning when the children were between 7.5-8 years old and 11 years old. Complete data on the SCAS-P were available at one or more time points for 238 participants. For participants who had complete data on the SCAS-P at two or more time points, a completed questionnaire from one of these time points was randomly selected in order to fully reflect the sampled age range; selection was randomized using the Excel RAND function. The mean age at time of completion was 8.9 years (SD = 1.1 years; see Table 3.1). The sex ratio (83.6% boys) approximates that observed in the general ASD population (Baio, 2014). Cognitive functioning data, collected when children were assessed between ages 8.5 - 9, was available for 212 (89%) of the participants and are reported solely for the purpose of sample description. Cognitive functioning ranged from superior abilities to severe intellectual impairment, with 77 (32.4%) children having a full-scale IQ < 70. As a standard IQ score could not be obtained for those participants whose cognitive abilities were lower than could be estimated using an age-appropriate scale, the standard score range is not reported here.
Table 3.1

*Sample Descriptive Statistics*

<table>
<thead>
<tr>
<th>Variable</th>
<th>N (%) or mean (SD)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Children</strong></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>199 (83.6%)</td>
</tr>
<tr>
<td>Female</td>
<td>39 (16.4%)</td>
</tr>
<tr>
<td>Age (years)</td>
<td>8.9 (1.1)</td>
</tr>
<tr>
<td>WISC-IV, verbal IQ Standard Score *</td>
<td>85.19 (18.2)</td>
</tr>
<tr>
<td><strong>Canadian Site</strong></td>
<td></td>
</tr>
<tr>
<td>Halifax, NS</td>
<td>40 (16.8%)</td>
</tr>
<tr>
<td>Montreal, QC</td>
<td>85 (35.7%)</td>
</tr>
<tr>
<td>Hamilton, ON</td>
<td>26 (10.9%)</td>
</tr>
<tr>
<td>Edmonton, AB</td>
<td>26 (10.9%)</td>
</tr>
<tr>
<td>Vancouver, BC</td>
<td>61 (25.6%)</td>
</tr>
</tbody>
</table>

Measures

This study included the SCAS-P, and the Child Behavior Checklist, 6-18 (CBCL; Achenbach & Rescorla, 2001), which was used to examine convergent and discriminant validity. The Wechsler Intelligence Scales for Children – 4th Edition (WISC-IV; Wechsler, 2003) and the Merrill-Palmer-Revised Scales of Development (M-P-R; Roid & Sampers, 2004) were used to assess cognitive functioning.

**Spence Children’s Anxiety Scale – Parent Form.** (SCAS-P; Spence, 1999). The SCAS-P is a freely available 38-item rating scale of anxiety symptoms in children ages 6-18 (Spence, 1999). Items are rated on their perceived frequency using a 0 (never) to 3 (always) scale. It was adapted from the child-report version of the SCAS and normative means have been reported based on typically-developing community and anxiety-disordered samples (Nauta et al., 2004). The SCAS-P was initially evaluated in Dutch and Australian community and clinical samples (Nauta et al., 2004), and in subsequent studies across a range of countries and cultural groups (Arendt et al., 2014; Ishikawa et al., 2013; Li et al., 2011; Orbay & Ayvasik, 2006; Whiteside & Brown, 2008). Most studies of the SCAS-P’s internal structure have reported a six-factor structure, reflecting correlated, though distinct, anxiety symptom groupings. These groupings of items within the SCAS-P are consistent with the anxiety disorder subtypes classified in the *Diagnostic and Statistical Manual of Mental Disorders* (4th ed., text. rev; *DSM-IV-TR*; American Psychiatric Association, 2000): panic disorder and agoraphobia, separation anxiety disorder, social phobia, specific phobia, OCD, and generalized anxiety disorder (GAD; Arendt et al., 2014; Ishikawa et al., 2013; Li et al., 2011; Nauta et al., 2004; Orbay & Ayvasik, 2006).
In addition to structural validity in samples of typically-developing children, the SCAS-P showed good to excellent internal consistency across the total score and subscale scores (Brown-Jacobsen, Wallace, & Whiteside, 2011; Nauta et al., 2004; Russell & Sofronoff, 2005; Whiteside & Brown, 2008). The total score significantly predicted an anxiety diagnosis in a clinical sample of 88 children (Brown-Jacobsen et al., 2011). The scale also showed good discriminant validity between the mean subscale scores and total score when compared across North American community and clinical samples of 85 children aged 9 to 18 (Whiteside & Brown, 2008).

Child Behavior Checklist, 6-18. (CBCL; Achenbach & Rescorla, 2001). The CBCL is a 113-item measure that asks parents to rate, on a three-point Likert scale, the frequency with which their child displays emotional and behavioural problems. The CBCL contains two broad scales reflecting internalizing and externalizing symptoms, and eight syndrome scales. Three syndrome scales are subsumed under the Internalizing scale: Anxious/Depressed, Withdrawn/Depressed, and Somatic Complaints. Two syndrome scales are subsumed under the Externalizing scale: Rule Breaking Behavior and Aggressive Behavior. The three remaining syndrome subscales — Social Problems, Thought Problems, and Attention Problems — loaded onto both the Internalizing and Externalizing domains in the original Achenbach and Rescorla (2001) analyses. In addition, the CBCL also includes six DSM-IV-TR oriented scales relating to Affective Problems, Anxiety Problems, Somatic Problems, ADHD, Oppositional Defiant Problems, and Conduct Problems (Achenbach & Rescorla, 2001).

The psychometric properties of the CBCL have been examined in children with ASD. The internal structure, scale reliability, criterion validity, and diagnostic accuracy of the
CBCL 6-18 were reported based on a sample of 122 children with ASD, ages 6-18, ranging in cognitive functioning (Pandolfi, Magyar, & Dill 2012). Their findings supported the unidimensionality of most CBCL syndrome scales, with the exception of the Thought Problems and Aggressive Behavior scales. The scale reliabilities were acceptable, ranging from $\alpha = .69$ to .94. Confirmatory factor analysis also supported the two-factor Internalizing-Externalizing structure. In addition, scores on the Anxious/Depressed, Withdrawn/Depressed, Somatic Complaints, Thought Problems, Internalizing Domain and Total Problems scales discriminated between children with and without a diagnosed concurrent psychiatric disorder.


Merrill-Palmer-Revised Scales of Development. (M-P-R; Roid & Sampers, 2004). The M-P-R is an individually administered measure of cognitive functioning. The M-P-R is appropriate for children age 2 months to 6.5 years, but was used in the present study when a child did not achieve a basal score on the WISC-IV. Children who were assessed using the M-P-R (at age 8.5 to 9 years) were described as having a verbal IQ < 70.

Data Analysis

Psychometric evidence to support the interpretation of the SCAS-P scores as indicators of anxiety severity in children with ASD was evaluated in several ways. First, the internal structure of the SCAS-P was evaluated through a CFA of the previously reported six-correlated-factor model (Arendt et al., 2014; Ishikawa et al., 2013; Li et al., 2011; Nauta et al., 2004; Orbay & Ayvasik, 2006). Given the small sample size relative to the number of
indicators and hypothesized factors (Brown, 2006), the examination of the original model was considered as an omnibus test, which could not fully discern areas of inadequate fit. Subsequent CFA with the Generalized Anxiety Disorder, Separation Anxiety Disorder, Social Phobia, Personal Injury Fears and separated Panic Disorder and Agoraphobia subscales allowed for an assessment of model fit in this sample, as well as the relationships among item scores within each of the subscales. Confirmatory factor analyses were conducted using the lavaan R package (Rosseel, 2012). Items were considered to be continuous for the 6-correlated factor model so as not to run into convergence problems, but were treated as categorical for individual subscales thereafter. The robust weighted least squares estimator was used for the CFA due to its appropriateness for smaller sample sizes, ordinal responding and for cases of serious violations of multivariate normality (Brown, 2006).

Each model’s fit was assessed through (a) inspection of the residual matrices for extreme outliers that could indicate localized areas of ill fit, (b) examination of the magnitude (> 0.32; Tabachnick & Fidell, 2001) and significance of standardized factor loadings, and (c) goodness of fit statistics (Brown, 2006). Hu and Bentler (1999) recommended the following combinational cut-off criteria for goodness of fit statistics to reduce Type II error rates: root mean squared error of approximation (RMSEA; <0.06), comparative fit index (CFI; >0.95), and Tucker-Lewis Index (TLI; >0.95). The 90% RMSEA confidence interval is reported to give a sense of uncertainty, though the decision was made on the point estimate (Hu & Bentler, 1999). Robust weighted least squares estimation produces a mean and variance adjusted $\chi^2$ test statistic which was not appropriate to the six-factor model due to deviation from multivariate normality, but was considered for individual subscale analyses (Hooper,
Coughlan, & Mullen, 2008). If subscales evidenced good fit, their internal consistency, convergent and discriminant validity were evaluated. Individual subscales that did not show good fit were removed from further analyses.

**Missing Data**

Of the parents who returned the SCAS-P ($n = 250$), 12 parents (4.8%) did not complete all 38 items. There were, at most, four missing responses for any item (< 2%); given these low rates, listwise deletion was considered appropriate. Although listwise deletion has the disadvantage of decreasing statistical power and producing larger standard errors when a considerable proportion of the sample is removed, it is an effective strategy for categorical data when it is missing completely at random and only a very small part of the sample is removed (Brown, 2006).

**Internal consistency.** As SCAS-P items are scored on an ordinal scale the internal consistency of each subscale was assessed using ordinal coefficient alpha, rather than coefficient alpha, which can be artificially deflated when used with Likert-type response scales (Zumbo, Gadermann, & Zeisser, 2007).

**Convergent and discriminant validity.** Further evidence to support the validity of SCAS-P subscales was sought through examination of their convergent and discriminant validity by correlations with CBCL subscales completed concurrent to the selected SCAS-P, using Pearson’s $r$. The CBCL subscales reflecting internalizing problems, including anxiety and depression (i.e., Internalizing problems, Anxious/Depressed, Anxiety Problems) as well as the subscales reflecting phenomenologically distinct symptoms of externalizing problems (i.e., Rule Breaking Behavior, Aggressive Behavior) were used in analyses.
Results

Confirmatory Factor Analyses

Evaluation of the six-correlated-factor model showed that although it met criteria based on the RMSEA = .03, none of the other criteria showed adequate fit (CFI = 0.80, TLI = 0.79, SRMR = 0.09). Inspection of each residual correlation matrix showed several values equal to or exceeding |0.07| suggesting localized points of ill fit in the solution. The three highest absolute residual values were 0.17 (items 21 and 27), 0.18 (items 38 and 25), and 0.19 (items 16 and 21). The Panic Disorder, Agoraphobia, Social Phobia, Personal Injury Fears, Separation Anxiety Disorder, and Generalized Anxiety Disorder factors were then evaluated individually. Of these six subscales, only the Social Phobia and Personal Injury Fears subscales showed poor fit, based on the indices of fit (see Table 3.2), and extreme absolute residual values (Personal Injury Fears: 0.10 [items 2 and 16]; 0.12 [items 16 and 21]); (Social Phobia: 0.21 [items 6 and 31]; 0.27 [items 6 and 26]). The Social Phobia and Personal Injury Fears subscales were removed from further analyses. Although no fit indices could be calculated for the Agoraphobia subscale because it only contained three items, it was also accepted based on large magnitude (>0.4) of all standardized factor loadings. The remaining three subscales showed good fit based on residual values, significant standardized factor loadings exceeding 0.42 (see Table 3.3), as well as $\chi^2$, CFI, TLI and RMSEA criteria.
Table 3.2

Comparison of Goodness of Fit Indices across Six-correlated Factor Model, and Individual Subscales

<table>
<thead>
<tr>
<th>Model</th>
<th>$\chi^2$ (df)</th>
<th>CFI</th>
<th>TLI</th>
<th>RMSEA (90% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Six-correlated factors</td>
<td>N/A</td>
<td>0.80</td>
<td>0.79</td>
<td>0.03 (0.02 0.04)</td>
</tr>
<tr>
<td>Generalized Anxiety</td>
<td>9.73 (9)</td>
<td>0.99</td>
<td>0.99</td>
<td>0.02 (0.00 0.08)</td>
</tr>
<tr>
<td>Separation Anxiety</td>
<td>14.45 (9)</td>
<td>0.98</td>
<td>0.97</td>
<td>0.05 (0.00 0.96)</td>
</tr>
<tr>
<td>Panic</td>
<td>6.71 (5)</td>
<td>0.99</td>
<td>0.97</td>
<td>0.04 (0.00 0.10)</td>
</tr>
<tr>
<td>Social Phobia</td>
<td>52.38 (9)**</td>
<td>0.95</td>
<td>0.92</td>
<td>0.14 (0.11 0.18)</td>
</tr>
<tr>
<td>Personal Injury Fears</td>
<td>12.64 (5)*</td>
<td>0.93</td>
<td>0.86</td>
<td>0.08 (0.02 0.13)</td>
</tr>
</tbody>
</table>

*Note. CFI = Comparative fit index; TLI = Tucker Lewis index; RMSEA = Root mean square error of approximation; CI = Confidence Interval; N/A = not applicable. All models were calculated using robust weighted least squares estimation. Data was treated continuously for the six-correlated factor model, and categorically for the remaining subscales. Numbers in bold meet the criteria set for good model fit. ** $p < .005$
Table 3.3

*Factor Loadings of Subscales Showing Good Fit*

<table>
<thead>
<tr>
<th>Generalized Anxiety</th>
<th>Factor loading</th>
</tr>
</thead>
<tbody>
<tr>
<td>1  Worries about things</td>
<td>.79**</td>
</tr>
<tr>
<td>3  Complains of stomach aches</td>
<td>.44**</td>
</tr>
<tr>
<td>4  Complains of feeling afraid</td>
<td>.85**</td>
</tr>
<tr>
<td>18 When my child has a problem, s(he) complains of his/her heart beating really fast</td>
<td>.62**</td>
</tr>
<tr>
<td>20  Worry that something bad will happen to him/herself</td>
<td>.74**</td>
</tr>
<tr>
<td>22  Child feels shaky when there is a problem</td>
<td>.48**</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Separation Anxiety</th>
<th>Factor loading</th>
</tr>
</thead>
<tbody>
<tr>
<td>5  Fear of being alone at home</td>
<td>.63**</td>
</tr>
<tr>
<td>8  Fear of being separated from caregivers</td>
<td>.67**</td>
</tr>
<tr>
<td>11  Worry about bad things happening to family members</td>
<td>.46**</td>
</tr>
<tr>
<td>14  Fear of sleeping alone</td>
<td>.63**</td>
</tr>
<tr>
<td>15  Trouble going to school in the mornings due to fear or nervousness</td>
<td>.56**</td>
</tr>
<tr>
<td>38  Child fears staying away from home overnight</td>
<td>.67**</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Panic</th>
<th>Factor loading</th>
</tr>
</thead>
<tbody>
<tr>
<td>12 Complains of shortness of breath</td>
<td>.56**</td>
</tr>
<tr>
<td>19  Child starts to tremble or shake without warning</td>
<td>.90**</td>
</tr>
<tr>
<td>28  Child suddenly feels scared for no apparent reason</td>
<td>.63**</td>
</tr>
<tr>
<td>30  Complains of suddenly becoming dizzy or faint for no apparent reason</td>
<td>.99**</td>
</tr>
<tr>
<td>32  Complains heart suddenly beating too quickly</td>
<td>.66**</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Agoraphobia</th>
<th>Factor loading</th>
</tr>
</thead>
<tbody>
<tr>
<td>25  Fear of travelling in a car, bus or train</td>
<td>.70**</td>
</tr>
<tr>
<td>27  Fear of being in crowded places</td>
<td>.94**</td>
</tr>
<tr>
<td>34  Fear of being in small closed places</td>
<td>.52**</td>
</tr>
</tbody>
</table>

*Note.* Item wording is paraphrased. ** Loading is significant at the .005 level.

The Generalized Anxiety, Separation Anxiety, Panic and Agoraphobia subscales showed small to large correlations with one another (see Table 3.4). The lowest correlations were found between the Separation Anxiety and Panic, and Agoraphobia subscales, while the
highest correlations were between the Agoraphobia, Panic and Generalized Anxiety subscales.

Table 3.4

*Correlations between Subscales*

<table>
<thead>
<tr>
<th>Subscale</th>
<th>Generalized Anxiety</th>
<th>Separation Anxiety</th>
<th>Panic</th>
</tr>
</thead>
<tbody>
<tr>
<td>Generalized Anxiety</td>
<td>--</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Separation Anxiety</td>
<td>.56**</td>
<td>--</td>
<td></td>
</tr>
<tr>
<td>Panic</td>
<td>.78**</td>
<td>.27**</td>
<td>--</td>
</tr>
<tr>
<td>Agoraphobia</td>
<td>.89**</td>
<td>.31**</td>
<td>.87**</td>
</tr>
</tbody>
</table>

*Note.** $p < .005$

**Internal Consistency**

The internal consistency of all subscales was above the minimal acceptable level ($\geq 0.65$; DeVellis, 2003): Generalized Anxiety Subscale ordinal $\alpha = .81$; Separation Anxiety Subscale ordinal $\alpha = 0.77$; Panic subscale ordinal $\alpha = 0.80$; Agoraphobia subscale ordinal $\alpha = 0.75$.

**Convergent and Discriminant Validity**

Correlations between the SCAS-P Generalized, Separation, Panic and Agoraphobia subscales were of medium to large magnitude (Cohen, 1992) with the CBCL Anxiety Problems subscale, offering support of convergent validity. There was less evidence for convergence with the Anxious/Depressed and Internalizing Problems subscales, with correlations of small to medium magnitude observed. Correlations were generally lower
(<0.34) with the Aggressive Behavior, Externalizing Problems and Rule-Breaking Behavior subscales, supporting their discriminant validity. One exception, however, was a medium correlation between the Agoraphobia and Aggressive Behaviour subscales. Table 3.5 presents all correlations between the SCAS-P subscales and the relevant CBCL subscales.

Table 3.5

Convergent and Discriminant Validity of Identified SCAS-P Subscales with CBCL Subscales

<table>
<thead>
<tr>
<th>CBCL subscale</th>
<th>Generalized Anxiety (Pearson’s r)</th>
<th>Separation Anxiety (Pearson’s r)</th>
<th>Panic (Pearson’s r)</th>
<th>Agoraphobia (Pearson’s r)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Anxiety Problems</td>
<td>.57**</td>
<td>.54**</td>
<td>.41**</td>
<td>.49**</td>
</tr>
<tr>
<td>Anxious/Depressed</td>
<td>.39**</td>
<td>.26**</td>
<td>.23**</td>
<td>.29**</td>
</tr>
<tr>
<td>Internalizing Problems</td>
<td>.39**</td>
<td>.29**</td>
<td>.32**</td>
<td>.37**</td>
</tr>
<tr>
<td>Aggressive Behavior</td>
<td>.21**</td>
<td>.34**</td>
<td>.33**</td>
<td>.41**</td>
</tr>
<tr>
<td>Externalizing Problems</td>
<td>.19**</td>
<td>.21**</td>
<td>.28**</td>
<td>.34**</td>
</tr>
<tr>
<td>Rule-Breaking Behavior</td>
<td>.14*</td>
<td>.26**</td>
<td>.22**</td>
<td>.29**</td>
</tr>
</tbody>
</table>

Note. CBCL = Child Behavior Checklist 6-18 (Achenbach & Rescorla, 2011). ** p < .005 * p < .05

Summary of Results

The results of the current study suggest that the previously reported factor structure of the SCAS-P is not a good fit for a sample of 7.5- to 11-year-olds with ASD. Examination of each SCAS-P subscale and item groupings that reflected DSM-IV-TR and Diagnostic and Statistical Manual of Mental Disorders (5th Ed.; DSM-5; APA, 2013), anxiety disorder symptom categories (panic disorder and agoraphobia, panic disorder, agoraphobia, separation anxiety disorder, social phobia, specific phobia, and GAD) revealed that the Generalized
Anxiety, Separation Anxiety, Agoraphobia and Panic subscales were supported. These four subscales each showed acceptable internal consistency and were strongly correlated with one another. All subscales also showed good convergent validity with the Anxiety Problems subscale of the CBCL, though lower correlations with the Anxious/Depressed and Internalizing Problems subscales. All four subscales showed lower correlations with the Rule-Breaking Behavior, Aggressive Behavior subscales, and Externalizing Problems factor, suggesting good discriminant validity. The Agoraphobia subscale showed a medium correlation with the Aggressive Behaviour subscale of the CBCL, suggesting weaker evidence of discriminant validity.
Chapter 4: Study 2

Anxiety disorders are highly comorbid with autism spectrum disorders (ASD; van Steensel et al., 2011). However, it is unknown what predicts anxiety development in this population. Research in typically-developing children has identified several factors that increase a child’s probability of developing clinically significant anxiety. First, the expression of disordered in typically-developing children changes with age (Bongers et al., 2003; Kessler et al., 2012; van Oort et al., 2009). Developmentally, childhood and adolescence are risk periods for the onset of anxiety disorders. Fifty percent of anxiety disorder diagnoses become apparent between age six and 21, and the odds of developing an anxiety disorder later in life decrease substantially (Kessler, Chiu, & Demler, 2005). In addition to their restricted age of onset, most anxiety disorders present more commonly in females (McLean & Anderson, 2009). Anxiety is also highly influenced by both hereditary and environmental family factors. Genetics account for about 30% of the variance in childhood anxiety disorders (Gregory & Eley, 2007), though the degree of heritability is higher for girls than boys (Eaves et al., 1997; Feigon et al., 2001) and also depends on the age of onset, the specific anxiety disorder, and the severity or facet of anxiety in question (Gregory & Eley, 2011). Shared environments (e.g., family factors, socioeconomic status) account for an additional 20% of the variance and unique environmental influences account for the remaining 50% (Gregory & Eley, 2007). Lastly, anxiety disorders in children are also predicted by a behaviourally inhibited temperament in early childhood, one that can include emotional negativity, withdrawn or cautious responding to novelty, and extreme social reticence (Degnan et al., 2010). This literature offers some starting points for evaluating whether the above individual and family factors have predictive power with regard to anxiety disorders in children with ASD.
Predictors and Correlates of Anxiety Development in Autism Spectrum Disorder

**Gender differences.** Due to the strong evidence that females in the general population are at higher risk for most anxiety disorders, sex differences in anxiety in children with ASD have received some focus. However, the majority of studies have reported no differences, albeit often based on smaller sample sizes of females ($n = 14 – 28$) due to the high male-to-female ratio in ASD (Gotham et al., 2015; May et al., 2014; Park et al., 2012; Simonoff et al., 2008; Solomon et al., 2011), or based on broad measures of anxiety or internalizing symptoms (Gotham et al., 2013; Park et al., 2012; Simonoff et al., 2008). An exception to this general conclusion was reported by May et al. (2014), who examined multiple anxiety domains and found higher social anxiety symptoms in 28 females relative to 28 males, all with IQ $> 70$, though no differences in other domains. Additionally, Gotham et al. (2015) found that sex interacted with age, whereby males with ASD showed higher general anxiety levels than females at late school age, but females showed steeper symptom increases during adolescence, and no differences were apparent by young adulthood. Examining gender differences in samples with higher numbers of females and across specific anxiety disorders is thus an important area for research.

**Family factors.** Due to the strong evidence that elevated anxiety in the general population runs in families (Rapee, 2011), some research has also been conducted on the rates of internalizing psychopathology in the parents or extended family of children with ASD. Retrospective rates of internalizing disorders were reported to be higher in mothers, first-, second- and third-degree relatives of individuals with ASD relative to both controls (Micali et al., 2004) and relatives of individuals with Down Syndrome (Bolton et al., 1998). Cross-sectional studies specific to the incidence of anxiety disorders have reported a similar
pattern: rates of social anxiety symptoms were found to be higher in mothers, though not fathers, of children with ASD relative to controls (Kuusikko-Gauffin et al., 2012). Similarly, rates of anxiety disorders were equally high in twins of children with ASD whose profiles were consistent with the broader autism phenotype, and higher than controls even in unaffected co-twins (Hallett, Ronald, et al., 2013). Taken together, these studies suggest that there may be some genetic commonalities between ASD and anxiety disorders. However, there is not sufficient evidence to suggest that anxiety in ASD is hereditary. Whether symptoms of internalizing disorders in family members are related to their incidence in children with ASD remains to be determined. Longitudinal research is needed to better understand the causal relationship of parental psychopathology to anxiety in ASD.

**Verbal and intellectual abilities.** There has been some suggestion that elevated anxiety is more prevalent at the higher-functioning end of the autism spectrum (White et al., 2009). This has been tested empirically across various studies that have measured anxiety symptoms in individuals across levels of verbal ability, adaptive functioning, and IQ, but results have been inconsistent. Empirical studies that have addressed this question have shown mixed findings. Based on various anxiety rating scales, five studies with ASD samples ranging from 172 to 1202 have reported higher parent-rated anxiety symptoms in higher-functioning children (Hallett, Lecavalier, et al., 2013; Lecavalier, 2006; Mazefsky et al., 2010; Sukhodolsky et al., 2008; Weisbrot et al., 2005). Specifically, in a sample of 75 children (ages 6-12), those with IQ scores above 70 were more likely to have high levels of anxiety as rated by parents and teachers on the anxiety scale of the Child and Adolescent Symptom Inventory (CASI-Anxiety; Weisbrot et al., 2005). In addition, a positive correlation between IQ and anxiety on the CASI-Anxiety was also found in a study of 171
children, ages 5-17 who had high levels of externalizing behaviours (e.g., hyperactivity, tantrums, aggression, self-injury) in addition to ASD (Sukhodolsky et al., 2008). The children in this sample with IQ scores above 70 were also more likely to meet CASI-Anxiety cut-off criteria for any anxiety disorder than children with IQs below 70 (Sukhodolsky et al., 2008). Similarly, a small but significant positive correlation was reported between parent-reported symptoms based on the CBCL anxiety/depressed subscale and IQ in a sample of 1,202 children with ASD who ranged age from 4-17 (Mazurek & Kanne, 2010).

Higher rates of anxiety were also associated with higher adaptive functioning in a sample of 353 children ages 3-21 receiving educational services for ASD (Lecavalier, 2006). A strength of this study was its reliance on the Anxious/Insecure subscale of the Nisonger Child Behavior Rating Form (Aman, Tassé, Rojahn, & Hammer, 1996). This scale was specifically designed to assess behavioural and emotional problems in children with intellectual disabilities, and has shown good construct validity in an ASD sample (Lecavalier, Aman, Hammer, Stoica, & Mathews, 2004).

Hallett, Lecavalier and coauthors (2013) presented the most thorough examination of whether higher anxiety was due to higher verbal ability rather than overall IQ alone. In their sample of 415 children (ages 4-17), the average anxiety level on the parent-rated CASI-Anxiety for children with IQ scores of 70 or above was significantly higher than that of children with IQs below 70, and high anxiety was also related to being verbal (Hallett, Lecavalier, et al., 2013). Hallett and coauthors (2013) also reported that items requiring a high degree of verbal ability (e.g., “has nightmares about being separated from parents”) were rarely endorsed across all levels of cognitive functioning. Parents of children with IQ scores lower than 70 were also less likely to endorse items referring to “worries” or
“complaints.” Moreover, anxiety levels remained significantly higher in children with IQ scores above 70 even when verbally endorsed items were removed from the analysis, suggesting that more observable expressions of anxiety may be more frequent in higher functioning children (Hallett, Lecavalier, et al., 2013).

Other studies have not found a relationship between cognitive functioning and levels of anxiety in individuals with ASD. Simonoff and coauthors (2008) reported no correlation between IQ and likelihood of an anxiety disorder diagnosis, based on the Child and Adolescent Psychiatric Assessment structured parent interview (CAPA; Angold & Costello, 2000) in their sample of 112 children who ranged in IQ from 19-124. Similarly, no correlation between parent-rated anxiety on the anxiety subscale of the Child Behavior Checklist (CBCL; Achenbach & Rescorla, 2001) was found in a sample of 134 children with ASD who ranged in IQ from 48-124 (Eussen et al., 2013). Notably, there were few participants at either extreme end of the distribution in cognitive functioning, which may have accounted for the finding (Eussen et al., 2013). Finally, in a sample of 95 high-functioning children (ages 6-18; IQ≥70), no difference in IQ was found between children with elevated and average anxiety based on parent report on the CBCL (Strang et al., 2011).

The mixed findings in the afore-mentioned studies may be due to several factors. This includes the statistical analysis used (e.g., correlation: Degnan et al., 2010; Mazurek & Kanne, 2010; Sukhodolsky et al., 2008; versus comparison of mean anxiety level across low and high-functioning groups: Lecavalier, 2006); the specificity of parent-report rating scales (e.g., anxiety/depression subscale of the CBCL: Eussen et al., 2013; Fox et al., 2005; Mazurek & Kanne, 2010 versus the CASI-Anxiety scale: Hallett, Lecavalier, et al., 2013; Weisbrot et al., 2005); the types of measurement tools used to measure anxiety (e.g., rating
scales: Chronis-Tuscano et al., 2009; Hirshfeld-Becker et al., 2007; Mazurek & Kanne, 2010; Muris et al., 2011; Strang et al., 2011; Sukhodolsky et al., 2008 versus structured interviews: Simonoff et al., 2008); the scope of anxiety measured (e.g., general anxiety: Mazurek & Kanne, 2010; Strang et al., 2011 versus specific anxiety disorder symptoms: Sukhodolsky et al., 2008; versus the presence of a specific anxiety disorder diagnosis: Simonoff et al., 2008); and other sample characteristics (e.g., externalizing behaviours: Sukhodolsky et al., 2008). These differences across studies make drawing clear conclusions difficult.

Further complicating this discussion are suggestions that anxiety in minimally verbal individuals with ASD may appear differently on the surface than in children with well-developed language. Specifically, anxiety in children with poorly developed language may also lead to problem behaviours, which are not captured by commonly available anxiety rating scales (Cohen, Yoo, Goodwin, & Moskowitz, 2011; Moskowitz et al., 2013). It is also possible that different types of anxiety symptoms are more common across different levels of language and cognitive ability. For example, children who are nonverbal, or show little language may be more likely to show fearful or avoidant responses to specific stimuli (Moskowitz et al., 2017), while children with higher language ability may be more likely to report general, more abstract worries (e.g., inferring the thoughts or judgements of others). Understanding the expression of anxiety across levels of language and cognitive ability is important for both treatment planning and conceptualization, and can guide our understanding around the role of anxiety in ASD (Kerns & Kendall, 2012).
Developmental Changes in Anxiety in ASD

Research on developmental risk periods for anxiety development in ASD has also been limited. Five studies have explicitly focused on developmental changes in anxiety in ASD (Bitsika & Sharpley, 2015; Davis et al., 2011; Gotham et al., 2015; Teh et al., 2017; Vasa et al., 2013). One study examined symptoms across the lifespan, from infancy to > 65 (Davis et al., 2011), while the others focused on smaller age ranges in childhood, adolescence (Bitsika & Sharpley, 2015; Teh et al., 2017; Vasa et al., 2013) as well as young adulthood (Gotham et al., 2015). Mixed findings were reported regarding the pattern of change between middle-childhood and adolescence, with some reporting peaks in middle childhood and declines into adolescence (Bitsika & Sharpley, 2015), while others reported a linear increase into adolescence (Gotham et al., 2015; Vasa et al., 2013). Teh and co-authors (2017) used the SCAS-P in its original format, and reported no mean change in total anxiety, or individual subscale scores in 54 children, ages from 5-17, ranging in cognitive ability, over 10-19 month follow-up periods. However, they found that 20% of the sample showed changes in severity level of anxiety across the two time points, based on normative data from typically-developing children (Nauta et al., 2004). Drawing conclusions from these studies is difficult due to differences in the age ranges examined, the instruments used, and the statistical analyses conducted. Nonetheless, the four cross-sectional studies indicated that anxiety ratings are lowest in early childhood and are dynamic over time (Bitsika & Sharpley, 2015; Davis et al., 2011; Gotham et al., 2015; Vasa et al., 2013). The only study reporting longitudinal data suggested that the amount of change varies by individual, though no change in mean anxiety levels (Teh et al., 2017). Whether anxiety ratings vary across individuals
during middle childhood specifically, and whether certain developmental periods are more susceptible to anxiety development, remains to be determined.

**Current Study**

The current study built on the limited research examining predictors of, and developmental changes in anxiety over time in the ASD population. Specifically, the study focused on the predictors, occurrence, and trajectories of anxiety development in middle childhood in this population. The current study measured parent and child predictive factors in early childhood and evaluated change and occurrence of parent-rated Generalized, Separation Anxiety, Panic and Agoraphobia symptoms over middle childhood. Specifically, the current study was guided by the following research questions about anxiety in children with ASD:

1. A) What is the level of parent-rated Generalized, Separation Anxiety, Panic and Agoraphobia symptoms experienced at each of ages 7.5-8, 8.5-9, 9.5-10, and 10.5-11? B) What proportion of children showed “Elevated” levels of parent-rated Generalized and Separation Anxiety based on SCAS-P norms?

2. What is the trajectory of parent-rated Generalized, Separation Anxiety, Panic disorder and Agoraphobia symptoms over middle childhood (age 7 to 11 years)?

3. Do A) language ability B) sex C) parent internalizing symptoms and D) parent-rated child anxiety in early childhood account for a significant amount of variance in the tendency towards parent-rated Generalized Anxiety, Separation Anxiety, Panic and Agoraphobia levels between ages 7-11?
Methods

Participants

The current study drew on existing data from Phase II of the multi-site, longitudinal Pathways in ASD study that follows children with ASD from age 2-19 (N = 308). Recruitment and data collection occurred across five sites (see Table 4.1). Children were diagnosed with ASD between ages 19 and 60 months (M = 39.23 months; SD = 8.76 months), and entered into the study an average of 1.5 months later. The Autism Diagnostic Observation Schedule (Lord et al., 2000) and the Autism Diagnostic Interview-Revised (Lord et al., 1994) were used in diagnostic evaluations. Children with identified genetic or chromosomal abnormalities, significantly impaired vision or hearing, cerebral palsy or other neuromotor disorders were excluded from the study. The study was approved by the Research Ethics Board at each site.

Data for the current study were collected at time of diagnosis and entry into the study (age 1.5-5 years of age), and then subsequently at four annual time points between 7.5 through 11 years of age. Participants whose parents had completed the rating scales of interest at study entry as well as one or more SCAS-P rating scales between ages 7.5-11 were included in the study (n = 262). Cognitive functioning data was available for 218/262 (83.2%) of the participants between 8.5-9 years of age. IQ scores ranged from superior functioning to severe intellectual impairment, with 88/218 (40.4%) participants scoring at an FSIQ below 70 on the WISC-IV, or being administered the M-P-R. The parent responding (i.e., the person most knowledgeable about the child) was most often the mother (91.7%). At time of entry into the study the mean age of parents responding was 36 (SD = 5.5 years), their median household income was $70,000 CAD, and the majority had received postsecondary education (89.5%).
Table 4.1

Sample Descriptive Statistics

<table>
<thead>
<tr>
<th>Variable</th>
<th>Mean (SD) or n (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Children</td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>220 (84.0%)</td>
</tr>
<tr>
<td>Female</td>
<td>42 (16.0%)</td>
</tr>
<tr>
<td>Age at diagnosis (months)</td>
<td>39.1 (8.76)</td>
</tr>
<tr>
<td>WISC-IV, FSIQ Standard Score *</td>
<td>84.71 (18.82)</td>
</tr>
<tr>
<td>Canadian Site</td>
<td></td>
</tr>
<tr>
<td>Halifax</td>
<td>41 (15.6%)</td>
</tr>
<tr>
<td>Montreal</td>
<td>96 (36.6%)</td>
</tr>
<tr>
<td>Hamilton</td>
<td>30 (11.5%)</td>
</tr>
<tr>
<td>Vancouver</td>
<td>61 (23.3%)</td>
</tr>
<tr>
<td>Edmonton</td>
<td>34 (13.0%)</td>
</tr>
</tbody>
</table>

Note. n = 262. WISC-IV = Wechsler Intelligence Scale for Children, Fourth Edition administered at age 8.5-9, (Wechsler, 2003) * available for n = 166 participants

Measures

Spence Children’s Anxiety Scale – Parent report. (SCAS-P; Spence, 1999). The SCAS-P is a 38-item questionnaire assessing anxiety symptoms in children ages 6-18. Items are rated on their perceived frequency using a 0 (never) to 3 (always) scale. The original factor structure of the SCAS-P included six factors reflecting panic attacks and agoraphobia, separation anxiety disorder, social phobia, physical injury fears, OCD, and GAD (Nauta et al., 2004). Study one demonstrated that the Generalized Anxiety, Separation Anxiety, Panic, and Agoraphobia factors were supported through confirmatory factor analysis in the Pathways in ASD cohort of children ages 7.5-11 (Jitlina et al., 2016). These subscales also showed acceptable evidence of convergent validity with the Anxiety Problems subscale of the Child Behaviour Checklist, for ages 6-18 (Achenbach & Rescorla, 2000; CBCL 6-18), though
lower convergence with the CBCL Anxious/Depressed and Internalizing Problems subscales. All four subscales also showed acceptable discriminant validity with the Externalizing Problems, and Rule-Breaking CBCL 6-18 subscales. The Generalized Anxiety, Separation Anxiety, Panic, and Agoraphobia subscales were thus used as the outcome measures in the current study. Standardized scores (T scores), and cut-offs indicating “Elevated” status are provided based on an aggregated sample across multiple studies for boys and girls ages 7-9, and 10-13 (Spence, 2017).

**Clinical Evaluation of Language Fundamentals, 4th Edition.** (CELF-4; Semel, Wiig, & Secord, 2003). The CELF-4 is a measurement tool designed to identify and diagnose language disorders in children ages 5–21. The Core Language composite standard score is based on subtests that assess oral comprehension, vocabulary, and syntax, and provides a measure of overall language performance. Composite scores on the CELF-4 show good evidence of inter-rater reliability, with scores ranging from 0.88-0.99. The CELF-4 also shows good internal consistency across subscales ($\alpha = 0.87-0.95$; Semel et al., 2003). The CELF-4 shows good specificity and sensitivity when discriminating between children with severe and average language abilities (Semel et al., 2003), although its utility in discriminating between children with severe language difficulties from those with only moderate difficulties has been questioned (Crowley, 2010). The CELF-4 manual reports the CELF-4 has good content validity based on review by an expert panel (Semel et al., 2003).

**Preschool Language Scale, 4th Edition.** (Zimmerman et al., 2012). The PLS-4 is an individually administered standardized test used to assess receptive and expressive language skills, and to identify children with language impairments. The PLS-4 provides raw scores, standard scores, percentile ranks and age equivalency scores in the areas of auditory
comprehension and expressive language for children from birth through to age 6 years 11 months. The auditory comprehension subscale assesses a child’s understanding of language and nonverbal listening skills. The expressive communication subscale assesses a child’s ability to verbally communicate with others. The PLS-4 Total Language Score encompasses both a child’s auditory comprehension and expressive language abilities. The test-retest reliability of the subscales ranged from 0.82 to 0.95, and from 0.90 to 0.97 for the Total Language Score (Zimmerman & Castilleja, 2005). For children ages 2-4 years, internal consistency coefficients ranged from 0.91-0.94 (Zimmerman et al., 2012). PLS-4 scores have also been shown to be sensitive to differences in language ability specifically in children with ASD (Zimmerman & Castilleja, 2005).

**Symptom Checklist-90-Revised.** (SCL-90-R; Derogatis, 1994). The SCL-90-R is a 90-item self-report questionnaire that assesses adult psychopathology across nine symptom domains, and was administered to one parent (91.7% mothers) at study entry. Each item asks the respondent to rate each symptom on a five-point scale (0-4) that ranges from “not at all” to “extremely.” The four symptom domains of interest to the current study were the depressive (13 items), obsessive-compulsive (10 items), anxiety (10 items), and phobic anxiety (7 items) domains. The depressive domain includes items about dysphoric mood, withdrawal from typical activities and interests, decrease in motivation, loss of energy, suicidal ideation, and feelings of hopelessness (Derogatis, 1994). The obsessive-compulsive domain includes items reflecting obsessive thoughts as well as compulsions that are experienced as intrusive, unwanted, and uncontrollable by the individual (Derogatis, 1994). The anxiety domain includes items reflecting behavioural, somatic, and cognitive symptoms of anxiety such as nervousness, trembling, muscle tension, and feelings of panic and fear.
Finally, the phobic anxiety domain reflects persistent symptoms of fear that lead to escape or avoidance in response to crowded places or enclosed public places (i.e., agoraphobia; Derogatis, 1994). With regard to the scale’s reliability, a study of the SCL-90-R with separate samples of adult psychiatric outpatients \( n = 103; n = 94 \) showed high internal consistency \( (\alpha = .82-.90) \) in the depression, obsessive-compulsive, anxiety, and phobic anxiety domains. In addition, based on these samples, these domains show good test-retest reliability following a one-week delay \( (r = .82-.90) \), with acceptable reliability following a 10-week delay \( (r = .70 -.80; \) Derogatis, Rickels, & Rock, 1976). With regard to the scale’s validity, early research comparing the SCL-90-R to other established multidimensional psychopathology scales reported extensively on its convergent validity with the Minnesota Multiphasic Personality Inventory (MMPI; Wiggins, 1969). All domains showed moderate to good convergent validity with the comparable subscales of the MMPI \( (r = .50-.75) \), except the obsessive-compulsive domain, which has no comparable subscale on the MMPI (Derogatis et al., 1976). A subsequent validation study of the SCL-90-R in a German clinical sample found the SCL-90-R depression and obsessive-compulsive domains to have moderate to high convergent validity to the Inventory of Interpersonal Problems \( (r = .61 \text{ and } r = .58, \text{ respectively}) \); while the anxiety \( (r = .45) \) and phobic anxiety domains showed weaker correlations \( (r = .45 \text{ and } r = .38, \text{ respectively}; \) Schmitz et al., 2000). In the same sample, the depression and anxiety domain scores independently showed acceptable concurrent validity with diagnostic classifications of depression and anxiety disorder classifications (Schmitz et al., 2000).

**Child Behavior Checklist.** (CBCL-1.5/5; Achenbach & Rescorla, 2001). The CBCL 1.5-5 is a rating scale of behaviours in children ages 18 months through 5 years of age.
Parents rate the applicability of 99 items that reflect externalizing and internalizing behaviours displayed by their child over the past two months. Each item is rated on a 3-point Likert scale (i.e., 0 = not true, 1 = somewhat true, 2 = very true or very often). The DSM-oriented Anxiety Problems (10 items) scale as of interest to the current study. The test-retest reliability of this scale was calculated on a group of 68 non-referred children over a period of 8 days, and the test-retest reliability was high (r = .85). Interparent agreement on the subscale was found to be acceptable (r = .66). All items of interest to the current study discriminate significantly between referred and non-referred children (Achenbach & Rescorla, 2001). Achenbach and Rescorla (2001) reported on the convergent validity of the Total Problems score of the CBCL 1.5/5 based on a sample of 36 referred, and 563 non-referred children. Convergent validity with several other parent-rated measures of problem behaviour in preschoolers showed correlations of Pearson’s r between .56 and .65. Evidence for the internal structure of the CBCL 1.5/5 in children with ASD comes from a study of 128 preschoolers with ASD (Pandolfi et al., 2009). Results of their confirmatory factor analysis suggested a good fit for all six subscales contributing to the Internalizing and Externalizing syndrome scales, though no analyses were reported for the DSM-oriented scales.

Wechsler Intelligence Scales for Children – 4th Edition. (WISC-IV; Wechsler, 2003). The WISC-IV is a norm-referenced measure of cognitive functioning intended for children ages 6-16 years of age. The full-scale IQ score captures functioning across four cognitive domains and is used for sample description purposes. The verbal IQ score reflects acquired verbal knowledge and verbal reasoning ability. Both scores are represented by a standard score (mean of 100; SD of 15).
**Merrill-Palmer-Revised Scales of Development.** (M-P-R; Roid & Sampers, 2004). The M-P-R is an individually administered measure of cognitive functioning. The M-P-R is appropriate for children age 2 months to 6.5 years, but was used in cases when a basal score was not achieved on the WISC-IV at age 8.5-9. Children who were assessed using the M-P-R were reported as having a FSIQ < 70 for the purpose of sample description. The M-P-R Developmental Index reflects the overall cognitive battery score, which captures a child’s cognitive, receptive language, memory, speed and visual-motor skills. Internal consistency estimates for the Developmental Index are strong ($\alpha = .97-.98$). The Developmental Index also shows a strong correlation with other cognitive assessment measures appropriate for early childhood include the Bayley Scales of Infant Development Mental Scale ($r = .92$), and the Stanford-Binet 5th Edition ($r = .80 - .86$; Roid & Sampers, 2004). The M-P-R Developmental Index Age Equivalent was used to guide cognitive ability estimation, as a standard score could not be calculated at the age of administration.

**Autism Diagnostic Observation Schedule.** (ADOS; Lord et al., 2000). The ADOS is a direct semi-structured assessment of communication, social interaction, and play for individuals suspected of having ASD. The assessment consists of 30-45 minutes of standardized activities that allow the examiner to make observations about the frequency of behaviours that are diagnostically relevant. Items are rated from 0 (nonautistic qualities) to 3 (severe autistic qualities) across the areas of social affect and restricted, repetitive behaviours based on the examiner’s behavioural observations. These ratings are used to formulate a score reflecting severity of ASD symptoms that allows for standardized comparison across the four modules, with scores of 1-3 considered to reflect little evidence for ASD symptoms (Gotham, Risi, Pickles, & Lord, 2007). The ADOS consists of four modules, one of which is
used depending on the developmental age and language level of the child at time of assessment. Module one is used for children over 31 months of age who are preverbal or spontaneously use single words. Module two is used for children who use phrase speech, but are not verbally fluent. Module three is used for verbally fluent children and adolescents. Module four is used for verbally fluent older adolescents or adults. The ADOS module selected at age 6 was the variable of interest to the current study, and thus psychometric properties of the ADOS are not reviewed here.

**Analyses**

Data were first examined to determine if there were any missing data patterns that suggested differences in age, sex of children, or IQ. Cohorts were also compared on IQ and anxiety scores to check for any differences. To approximate subgroups of children based on severity of language impairment, participants were classified into three subgroups. Subgroup classification was determined primarily based on language ability (i.e., PLS-4 Total Language Age Equivalent or CELF-4 Core Language Standard Score) at age 8.5-9. For children who did not complete language testing (n = 7) a subgroup was determined based on verbal reasoning ability (i.e., WISC-IV Verbal Intelligence Quotient; n = 4) or general cognitive ability at 8.5-9 (i.e., M-P-R Developmental Index Age Equivalent; n = 3). Finally, for the children (n = 24) who did not complete language or cognitive testing at age 8.5-9, subgrouping was based on the ADOS module administered at age 6, which is selected based on a child’s level of fluent speech. The cut-off scores used for the subgrouping are listed in Table 4.2. Thus, the subgroupings divided children into the “High” group, estimated to have developmentally-appropriate levels of language, the “Medium” group, estimated to be verbal, but with language abilities well-below those of peers, and the “Low” group, including
children who were nonverbal or showed extremely low levels of language.

Table 4.2

Language Impairment Subgroup Classification

<table>
<thead>
<tr>
<th>Group, sample</th>
<th>Decision Point</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>1</td>
</tr>
<tr>
<td></td>
<td>Language Ability; Verbal Reasoning Ability; ADOS Module;</td>
</tr>
<tr>
<td></td>
<td>age 8.5-9</td>
</tr>
<tr>
<td>Low, n = 65</td>
<td>PLS-4 A:E &lt; 4 years</td>
</tr>
<tr>
<td>Medium, n = 51</td>
<td>CELF-4 SS &lt; 70</td>
</tr>
<tr>
<td>High, n = 143</td>
<td>CELF-4 SS ≥ 70</td>
</tr>
</tbody>
</table>


Due to the high correlations between the depression, OCD, anxiety, and phobic anxiety domains of the SCL-90-R, treating predictors as independent of one another increased the possibility of missing significant parameter estimates. Thus, a principal components analysis was conducted to assess whether a higher-order latent component could account for the shared variance between SCL-90-R internalizing domains.

**Research question 1.** To examine the level of anxiety experienced by children at each of ages 7.5-8, 8.5-9, 9.5-10, and 10.5-11, mean anxiety levels and standard deviations at each time point were calculated. The distribution (skewness) of each outcome variable was examined. The proportion of males and females whose Generalized and Separation anxiety scores fell within the “Elevated” range based on ranges and T-Scores reported by Spence (2017) from an aggregate of community and clinical samples was also reported.
Research question 2. To examine the trajectory of anxiety outcomes over time, a multilevel modeling (Hox, 2010) approach was used. Multilevel modeling for longitudinal data considers repeated measurement occasions (Level 1) as nested within individuals (Level 2). This approach accounted for the shared variance in anxiety outcomes at each time point. For each child, change in anxiety was specified as a function of the child’s age, plus error. Linear change is captured by an intercept and a slope: the intercept reflects the mean level of anxiety when the child is seven years of age, and the slope reflects rate of change in parent-rated anxiety per year. All multilevel modeling analyses were conducted using the lme4 R package (Bates, Maechler, Bolker & Walker, 2017) using full maximum-likelihood estimation.

A series of multilevel models was constructed for each dependent variable: one for Generalized Anxiety, one for Separation Anxiety, Panic and Agoraphobia outcomes. Model 1 was an unconditional means model, which measured the proportion of variation in anxiety outcome scores between participants.

Model 1: Unconditional means model.

Level 1 (measurement occasion): $Y_{ij} = \pi_{0j} + r_{ij}$

Level 2 (child): $\pi_{0j} = \beta_{00} + u_{0j}$

Composite: $Y_{ij} = \beta_{00} + u_{0j} + r_{ij}$

At Level 1, the anxiety score at each measurement occasion $i$, within each child $j$ ($Y_{ij}$), was modeled as a function of the mean anxiety outcome score for each child $j$ ($\pi_{0j}$), as well as a residual term reflecting differences at each measurement point around the mean outcome score $j$ ($r_{ij}$). At Level 2, the mean anxiety outcome score for each child $j$ ($\pi_{0j}$) was modeled as a function of the mean of anxiety in the overall sample ($\beta_{00}$), plus a child-specific
deviation from the sample mean \((u_{0j})\). The intraclass correlation coefficient (ICC; \(\tau_{00}/(\tau_{00} + \sigma^2)\)), of the unconditional model was calculated to reflect the proportion of variability between children at Level 2, and to help determine the need for multilevel modeling.

In order to determine whether there was change in anxiety from age 7 to 11 (i.e., across measurement occasions), model 2, the unconditional growth model, included age at Level 1. Age was rescaled to intercept with zero at 7 years of age in order to facilitate interpretation. A quadratic \((\text{age}^2)\) trajectory was also examined, but a linear trajectory was pursued.

**Model 2: Unconditional growth model.**

Level 1 (measurement occasion): \(Y_{ij} = \pi_{0j} + \pi_{1j}(\text{age}_{ij}) + r_{ij}\)

Level 2 (child): \(\pi_{0j} = \beta_{00} + u_{0j}\)

\(\pi_{1j} = \beta_{10} + u_{1j}\)

Composite: \(Y_{ij} = \beta_{00} + u_{0j} + \beta_{10}(\text{age}_{ij}) + u_{1j}(\text{age}_{ij}) + r_{ij}\)

Comparisons in Level 1 variance estimates between the unconditional growth and unconditional models were conducted using the Pseudo \(R^2\) (Hayes, 2006), which compares the within-child variance of the unconditional growth model to that of the unconditional model: \(1 - (\sigma^2 \text{ of unconditional growth model} / \sigma^2 \text{ of unconditional model})\).

**Research question 3.** As there was no change in age slopes for Generalized, Separation Anxiety, Panic or Agoraphobia outcomes, or substantial variance in slopes across children, multiple linear regression was used for subsequent analyses. The outcome measures were considered the tendency towards Generalized Anxiety, Separation Anxiety, Panic or Agoraphobia between ages 7-11, calculated as the mean across the four time points in each domain. Child’s sex, and child’s language ability (i.e., High, Medium, Low), parent-
rated child anxiety symptoms in early childhood (CBCL Anxiety Problems), and parental internalizing symptoms (SCL-90-R composite score) in early childhood, were included in the model as predictors. Language ability was dummy coded. Parental internalizing symptoms and anxiety in early childhood variables were centered to the grand mean to facilitate interpretation.

**Results**

**Preliminary Analyses**

No significant differences across cohorts were apparent based on any of the four anxiety outcomes at any time point, or in IQ scores (based on the WISC-IV or M-P-R Developmental Index Raw Score). The distributions of IQ scores on both the WISC-IV and M-P-R and proportions of children receiving the M-P-R rather than the WISC-IV were comparable.

Table 4.3 includes an overview of the predictor variables examined including the means, standard deviations, ranges, and intercorrelations among the variables.
Table 4.3

*Intercorrelation Matrix, Means and Standard Deviations for Level 2 Predictor Variables*

<table>
<thead>
<tr>
<th>Predictor</th>
<th>Mean (SD)</th>
<th>Range</th>
<th>1.</th>
<th>2.</th>
<th>3.</th>
<th>4.</th>
<th>5.</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. CBCL 1.5/5</td>
<td>57.20 (8.77)</td>
<td>50-84</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Anxiety Problems</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>2. SCL-90-R</td>
<td>58.14 (10.31)</td>
<td>34-81</td>
<td>.25**</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Depression</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>3. SCL-90-R OCD</td>
<td>56.94 (11.38)</td>
<td>37-81</td>
<td>.29**</td>
<td>.81**</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Phobic Anxiety</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>4. SCL-90-R</td>
<td>49.49 (8.22)</td>
<td>44-76</td>
<td>.35**</td>
<td>.51**</td>
<td>.55**</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Anxiety</td>
<td></td>
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<td></td>
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</tr>
<tr>
<td>5. SCL-90-R</td>
<td>52.82 (11.19)</td>
<td>37-81</td>
<td>.32**</td>
<td>.79**</td>
<td>.76**</td>
<td>.57**</td>
<td></td>
</tr>
</tbody>
</table>

*Note.* CBCL = Child Behavior Checklist Ages 1.5-5; SCL-90-R = Symptom Checklist-90 Revised; OCD = Obsessive Compulsive Disorder; **p < 0.01

Results of a principal components analysis suggested that all four SCL-90-R internalizing domains loaded onto one component, which accounted for 75.25% of the variance between the individual domains (see Table 4.4). Similarly, visual inspection of the scree plot suggested a one-factor solution was the best fit for the data. Consequently, the SCL-90-R depression, OCD, anxiety, and phobic anxiety scores were summed to produce an internalizing composite score.
Table 4.4

*Component Loadings of Principal Components Analysis with SCL-90-R*

<table>
<thead>
<tr>
<th>Scale</th>
<th>Component Loading</th>
</tr>
</thead>
<tbody>
<tr>
<td>SCL-90-R Depression</td>
<td>.91</td>
</tr>
<tr>
<td>SCL-90-R OCD</td>
<td>.91</td>
</tr>
<tr>
<td>SCL-90-R Anxiety</td>
<td>.91</td>
</tr>
<tr>
<td>SCL-90-R Phobic Anxiety</td>
<td>.74</td>
</tr>
</tbody>
</table>

*Note.* SCL-90-R = Symptom Checklist-90 Revised; OCD = Obsessive Compulsive Disorder

**Missing Data**

There was missing data both at the level of the outcome and the predictor variables. Of the 308 children who entered Phase II of the larger study (i.e., data collection from ages 7.5-11), 262 had at least one returned SCAS-P questionnaire over the four collection time points. The number of completed SCAS-P questionnaires at each time point varied. In reviewing patterns in the missing data across time points, there was indication of drop out over time, missingness due to not yet having reached time point (due to younger cohorts in accelerated, multi-wave design) as well as intermittent missingness (individuals coming in and out of the study). Of the 262 children with one returned questionnaire, 236 had complete data on language or cognitive ability measures, CBCL 1.5/5 and SCL-90-R predictor variables. There were no differences between the group of 236 and 262 children according to sex, $\chi^2(1) = 2.3, p = .17$, age, $t(17.80) = -1.49, p = 0.15$ or cognitive ability at age 9.5-10 WISC IV FSIQ, $t(9.52) = -.07, p = 0.94$, M-P-R Developmental Raw Score $t(16.22) = 0.60, p = .56$.

**Results for research question 1.** The mean and standard deviation of the Generalized Anxiety, Separation Anxiety, Panic Disorder and Agoraphobia subscale scores
are reported in Table 4.5, and presented in box plots in Figures 4.1, 4.2 and 4.3. Because SCAS-P items have a restricted response range (0 - 3) and 0 scores were common, all scores showed floor effects (positive skew) at each time point, with the majority of children scoring in the lower end of each scale. A positive skew with minimal variability was particularly prominent for the panic disorder subscale (potential score range 0-20), where the median score was 0 at each time point, and mean scores ranged from 0.35-0.45.

Table 4.5

*Mean Scores and Standard Deviations of Outcome Variables Over Time*

<table>
<thead>
<tr>
<th>Score</th>
<th>7.5-8</th>
<th>8.5-9</th>
<th>9.5-10</th>
<th>10.5-11</th>
</tr>
</thead>
<tbody>
<tr>
<td>n</td>
<td>M (SD)</td>
<td>n</td>
<td>M (SD)</td>
<td>n</td>
</tr>
<tr>
<td>GAD</td>
<td>176</td>
<td>2.31 (1.95)</td>
<td>196</td>
<td>2.50 (2.17)</td>
</tr>
<tr>
<td>SEP</td>
<td>172</td>
<td>3.09 (2.83)</td>
<td>195</td>
<td>3.01 (3.43)</td>
</tr>
<tr>
<td>Panic</td>
<td>175</td>
<td>0.38 (0.83)</td>
<td>195</td>
<td>0.41 (0.86)</td>
</tr>
<tr>
<td>Agor</td>
<td>177</td>
<td>0.79 (1.26)</td>
<td>196</td>
<td>0.73 (1.13)</td>
</tr>
</tbody>
</table>

*Note.* All scores derived from the Spence Children’s Anxiety Scale – Parent report. GAD = Generalized Anxiety Disorder subscale; SEP = Separation Anxiety subscale; Panic = Panic Disorder subscale; Agor = Agoraphobia subscale
Figure 4.1. Box plots of Spence Children’s Anxiety Scale-Parent Report Generalized and Separation subscale raw scores across ages 7.5-11 years. Box plots represent the interquartile ranges of scores between 25th-75th percentiles. Upper and lower bars indicate minimum and maximum scores.
Figure 4.2. Box plot of Spence Children’s Anxiety Scale-Parent Report Panic subscale raw scores across ages 7.5-11 years. Box plots represent the interquartile ranges of scores between 25th-75th percentiles. Upper and lower bars indicate minimum and maximum scores.
Figure 4.3. Box plot of Spence Children’s Anxiety Scale-Parent Report Agoraphobia subscale raw scores across ages 7.5-11 years. Box plots represent the interquartile ranges of scores between 25th-75th percentiles. Upper and lower bars indicate minimum and maximum scores.

At each time point, a proportion of children’s scores fell within the “Elevated” range, based on SCAS-P norms for 7-9, and 10-13 year-old male and female children. Box plots based on T-Scores for males and females at each time point are presented in Figures 4.4 and 4.5. The number of participants and proportion of the sample falling into the “Elevated” range is reported in Appendix A.
Figure 4.4. Box plot of Spence Children’s Anxiety Scale-Parent Report Generalized subscale T-Scores across four time points. Box plots represent the interquartile ranges of scores between 25th-75th percentiles. Upper and lower bars indicate minimum and maximum scores.
Figure 4.5. Box plot of Spence Children’s Anxiety Scale-Parent Report Separation subscale T-Scores across four time points. Box plots represent the interquartile ranges of scores between 25\textsuperscript{th}-75\textsuperscript{th} percentiles. Upper and lower bars indicate minimum and maximum scores.

**Multilevel Analyses**

With regard to the multilevel analyses, residuals were visually inspected in order to examine whether assumptions of normality and linearity were met. This revealed that for the Generalized and Separation Anxiety unconditional growth models, normality of the Level 1 residuals and Level 2 residuals could be assumed. Visual inspection of the QQ plots of the Level 1 residuals for Agoraphobia and Panic symptom outcome unconditional growth models suggested some positive skew. Furthermore, some heteroskedasticity was observed based on the plot of residuals versus fitted values.

Analysis of the unconditional model suggested variability in anxiety scores across measurement occasions within children for Generalized Anxiety ($\sigma^2 = 1.76$), Separation
Anxiety ($\sigma^2 = 3.17$), Panic ($\sigma^2 = 0.48$) and Agoraphobia outcomes ($\sigma^2 = 0.44$). Variance in the average level of anxiety (i.e., the intercepts) between children was apparent for Generalized Anxiety ($\tau_{00} = 2.80$, $SD = 1.67$) and Separation Anxiety ($\tau_{00} = 6.36$, $SD = 2.52$), though less so for Panic ($\tau_{00} = 0.25$, $SD = 0.50$) and Agoraphobia outcomes ($\tau_{00} = 1.00$, $SD = 1.00$).

The heterogeneity between children for both outcome measures supported the need for multilevel modeling: the ICC indicated that 61% of the variability in Generalized Anxiety outcome scores, 67% in Separation Anxiety scores, 36% in Panic scores and 68% in Agoraphobia scores was due to individual differences between children.

**Results for research question 2.** Results of the unconditional growth model suggested that the rates of change over time between ages 7-11 were relatively flat. That is, age slopes did not differ significantly from 0 for Generalized ($\beta_{00} = 0.06$, $t(170.03) = 1.06$, $p = .22$), Separation Anxiety ($\beta_{00} = -0.07$, $t(174.33) = -0.88$, $p = .38$), Panic ($\beta_{00} = 0.001$, $t(158.8) = 0.03$, $p = .97$) and Agoraphobia outcomes ($\beta_{00} = -0.02$, $t(446.50) = -0.67$, $p = .50$). Indeed, age accounted for little additional variance within each child across measurement occasions, as indicated by the Pseudo $R^2$ value, which compares the within-child variance of the unconditional growth model: Generalized Anxiety: Pseudo $R^2 = 1 - (1.34/1.76) = 0.24$; Separation Anxiety Pseudo $R^2 = 1 - (2.38/3.17) = 0.25$; Panic Pseudo $R^2 = 1 - (0.40/0.44) = 0.09$; Agoraphobia Pseudo $R^2 = 1 - (0.48/0.48) = 0$. Moreover, there was little variance between children in age slopes for Generalized ($r_{1i} = 0.25$, $SD = 0.50$), Panic ($r_{1i} = 0.02$, $SD = 0.15$) and Agoraphobia ($r_{1i} = 0.00$, $SD = 0.04$) outcomes. Some variance was observed in Separation Anxiety outcomes ($r_{1i} = 0.45$, $SD = 0.67$). Tables summarizing these results are available in Appendix B, C, D, and E.
Results for research question 3. Multiple linear regressions were calculated to predict a tendency towards parent-rated Generalized, Separation, Panic and Agoraphobia outcomes between ages 7-11 based on child’s sex, language ability, as well as parent-rated anxiety and parent internalizing symptomatology in early childhood.

Scatterplots of studentized residuals vs standardized predicted values were visually examined to assess for the assumptions of heteroskedasticity, independent errors and a linear relationship between independent and dependent variables. Clustered patterns were observed for the Panic and Agoraphobia outcome measures, suggesting that the assumptions of heteroskedasticity, a linear relationship and independence of errors were violated. Taken together, these analyses suggest that multiple linear regression analyses could not be reliably undertaken with the Panic and Agoraphobia outcomes. Heteroskedasticity could be assumed based on reasonable consistency of spread through the Generalized Anxiety and Separation Anxiety distributions. Further, visual inspection of scatterplots of the dependent vs. the Generalized and Separation Anxiety outcomes variables also suggested a linear relationship could be assumed. In order to check for multicollinearity, the correlation was examined. The correlation between the Early Childhood Anxiety and Parent Internalizing predictors ($r = .34, p < .005$) did not suggest that multicollinearity was a concern. Eleven multivariate outliers were identified using Mahalanobis distance (11.07) and Cook’s Distance > 1. Outliers all had a logical range of responses across predictors and outcome variables, and were thus not excluded from the analysis. The distribution of residuals was examined via histograms and P-P plots, and these indicated that normality of residuals could be assumed across the Generalized Anxiety and Separation Anxiety outcomes.
The regression equations were significant across all remaining outcomes: Generalized
\( F(5, 228) = 19.94, p < .005 \); Separation \( F(5, 229) = 10.40, p < .005 \). Based on the
adjusted \( R^2 \), the amount of variance accounted for was 16.7% for Separation Anxiety and
28.9% for Generalized Anxiety. Results specific to each predictive variable are described
below.

Language ability was a significant predictor of the tendency towards Generalized, and
Separation between ages 7-11. Specifically, children with average language abilities were
rated by their parents as having a greater tendency towards parent-rated Generalized and
Separation Anxiety than children who were nonverbal or showing very limited language
ability (Generalized Anxiety: \( \beta = 1.60, SE = 0.25, t(228) = 6.34, p < .005 \); Separation
Anxiety: \( \beta = 1.12, SE = 0.41, t(229) = 2.75, p = .006 \)). These results suggest that on average,
children with typical language ability were rated by their parents as having 1.6 raw score
points more Generalized Anxiety, and 1.12 raw score points more Separation Anxiety than
children who were nonverbal. There was also a significant difference in outcomes between
children with average language ability and children with below-average language abilities
(i.e., High vs. Medium groups) in the case of Generalized Anxiety (\( \beta = 0.74, SE = 0.27 \),
\( t(228) = 2.73, p = .007 \)), though not Separation Anxiety.

Sex was not a significant predictor of the tendency towards parent-rated Generalized
(\( \beta = 0.18, SE = 0.29 \), \( t(228) = 0.61, p = .55 \)) or Separation Anxiety(\( \beta = 0.49, SE = 0.46 \),
\( t(229) = 1.05, p = .30 \)) scores across ages 7-11.

Parent-rated anxiety problems in early childhood significantly predicted the tendency
towards anxiety outcomes in middle childhood: Generalized Anxiety (\( \beta = 0.07, SE = 0.013 \),
\( t(228) = 5.50, p < .005 \), and Separation Anxiety (\( \beta = -.11, SE = 0.02, t(229) = 5.46, p < .005 \)).
Specifically, for every one point T-Score increase in parent-rated anxiety in early childhood, there would be an associated 0.07 raw score increase in Generalized Anxiety, and a 0.11 raw score increase in Separation Anxiety. Parental internalizing symptoms significantly predicted the tendency towards parent-rated Generalized Anxiety ($\beta = 0.009$, $SE = 0.004$, $t(228) = 2.46$, $p = .015$), though not Separation Anxiety ($\beta = 0.006$, $SE = 0.006$, $t(229) = 0.98$, $p = .33$). Thus, for every one point T-Score increase in parent internalizing symptoms, raw score parent-ratings increased by 0.01 for Generalized Anxiety outcomes.
Chapter 5: Discussion

Anxiety in ASD merits research and clinical attention due to its high prevalence and deleterious impact on an individual’s functioning (van Steensel et al., 2011). However, research, assessment and treatment efforts are threatened by a lack of psychometrically sound and accessible tools for the measurement of anxiety, and the ambiguous definition of anxiety in ASD (Wood & Gadow, 2010). Furthermore, little is known about the development of anxiety in ASD, including its early predictors and changes in symptoms over time (Bitsika & Sharpley, 2015; Davis et al., 2011; Gotham et al., 2015; Vasa et al., 2013). This area of research is important for informing treatment and prevention research, and for improving the quality of life of individuals with ASD.

Chapter 5 includes a discussion of the findings from the two studies within this dissertation, which expanded on research on anxiety assessment and development in ASD. The research and clinical implications of the results, the limitations and strengths of each study as well as the dissertation overall are discussed.

Study 1

The current results suggest that the originally established structure of the SCAS-P is not a good fit for a sample of 7- to 11-year-olds with ASD, including children with intellectual impairment, and confirm a previous report (Magiati et al., 2016). These findings indicate the SCAS-P is not a valid and reliable tool for the assessment of anxiety in children with ASD. Examination of the internal structure of the SCAS-P is important given that this rating scale has been used as a primary anxiety measure in many studies of children with ASD (see Grondhuis & Aman, 2012; Wigham & McConachie, 2014 for reviews), and is freely available for clinical use in the community. The current results highlight a limitation of existing studies that have used the SCAS-P as a measurement tool in this population.
However, the Generalized, Separation, Panic and Agoraphobia subscales identified in the current study showed good psychometric properties and are promising subscales. Despite the limitations of the SCAS-P in measuring anxiety in ASD, previously collected SCAS-P data will benefit from re-examination using these four subscales.

A potential reason for the lack of fit of the original structure is the characteristics of ASD that impact anxiety presentation. Future research focused on adapting measures for ASD and on differences in the presentation of anxiety in ASD relative to the general population (e.g., a subdomain of anxiety related to insistence on sameness) will offer further clarification around the poor fit of most existing measures.

**Anxiety disorders in ASD.** Secondary analyses of SCAS-P subscales identified areas of misfit within the overall measure, led to identifying subscales suited for the ASD population and consistent with *DSM-5* anxiety disorder reclassification, and contributed to the overall understanding of anxiety phenomenology in ASD. First, the original Generalized Anxiety subscale was supported in this sample. Additionally, the factor’s internal consistency was acceptable (DeVellis, 2003), and consistent with a past report on the original GAD factor (Zainal et al., 2014). With regard to the nature of anxiety in ASD, these results are consistent with Hallett et al. (2013), who reported that a factor reflecting generalized anxiety was supported on the CASI-Anxiety in an ASD sample. Furthermore, support for a unique generalized anxiety factor is expected based on the research of Kerns et al. (2014, 2016) regarding distinctions between “traditional” and “atypical” anxiety in ASD. This suggests that generalized anxiety may present similarly across non-ASD and ASD populations.
The original Separation Anxiety Subscale was also supported in the current sample. This is consistent with several reports that have supported the presentation of separation anxiety in ASD. These have included anxiety measurement tools specifically revised or expanded for ASD in samples of children with high cognitive functioning (Kerns et al., 2014; Rodgers et al., 2016), as well as the CASI-Anxiety when used with a sample of children ranging in IQ (Hallett, et al., 2013). Finally, when the original Panic and Agoraphobia subscale was separated into two separate factors to reflect their re-classification in the DSM-5, both showed a good fit in the current sample.

Despite past reports of several parent rating scales including factors related to, or including, social anxiety (Hallett et al., 2013; Rodgers et al., 2016; White et al., 2014), the SCAS-P Social Phobia subscale was not supported in this sample. The lack of fit may be due to differences in the experience of social anxiety between children with and without ASD, or to a lack of specificity for social anxiety within the context of ASD symptoms. Of note, examination of extreme residual values suggested that item 6, fear of test taking, was an area of localized ill-fit in this subscale, though no extreme values were related to two other items regarding to school performance and attendance (items 10 and 15). Furthermore, there was no indication that the original factor reflecting specific phobias was a good fit for our sample. Given that prevalence estimates suggest specific phobia is the most common anxiety disorder in ASD (van Steensel et al., 2011), it is possible that the limited focus of the SCAS-P Personal Injury Fears subscale contributed to the misfit. Specifically, the Personal Injury Fears subscale items reflect the frequency with which the child experiences common childhood worries including fear of the dark, of dogs, the doctor or dentist, of heights, and of insects (Spence, 1999). Given that children with ASD have been reported to experience a
wide range of fears, including those that are both common (e.g., fears related to medical events; Evans et al., 2005) as well as extremely uncommon (e.g., fears of showers and flushing toilets; Jackson & King, 1982), this subscale may have missed a large proportion of the clinically significant and impairing fears presenting in children with ASD.

Taken together, examinations of anxiety subscales have suggested that DSM-5 anxiety diagnostic criteria may have some applicability in ASD, despite a lack of fit of the original factor structure of several existing anxiety rating scales (Hallett, et al., 2013; Sukhodolsky et al., 2008; White et al., 2014). Moreover, the current study suggests that existing data collected using the SCAS-P for children with ASD can offer valuable information if analyzed using the four individual subscales identified.

**Context.** The current study relied on a “top-down” approach, alongside other studies that have attempted to clarify the utility of several existing anxiety instruments for the ASD population (Hallett, Lecavalier, et al., 2013; Lecavalier et al., 2014; White et al., 2014; Wigham & McConachie, 2014). The lack of support for overall structural similarity in anxiety across ASD and non-ASD populations has clarified that, although the construct is not identical in the two groups, some subscales show viability. Indeed, some traditional conceptualizations of anxiety subtypes appear helpful for those with ASD. In contrast, fewer studies have used a “bottom-up” approach that attempted to capture the presentation of anxiety symptoms in ASD without relying on DSM typology. This approach has included analyses of qualitative data collected through focus groups with parents and children (Ozsivadjian et al., 2012), real-time recording of worrisome thoughts (Hare et al., 2015), as well as psychophysiological and neuroimaging studies of fear conditioning responses (Sterling et al., 2013; Top et al., 2016). A middle-ground approach was used by Kerns and
co-authors, who addressed the issue of assessment by first quantifying the commonalities and differences with anxiety presentation in the typically developing population (Kerns et al., 2014), and then adding to an existing interview to capture “atypical” symptom presentations, or those that do not fall neatly into DSM-5 criteria (Kerns et al., 2015). Through these varied approaches, this body of research is refining the diagnostic concept of anxiety in the ASD population.

**Study 2**

In Study 2, parent-rated Generalized, Separation, Agoraphobia and Panic symptoms in children with ASD were examined at four time points across middle childhood. When considered alongside previously reported rates of children with clinically significant anxiety, rates of children with “Elevated” Generalized Anxiety scores were largely comparable to the rate previously reported in a meta-analysis (15.4%; van Steensel et al., 2011), while rates of children with “Elevated” Separation Anxiety scores in this sample was higher (9%; van Steensel et al., 2011). Notably, a higher proportion of children were rated by parents as having Generalized and Separation Anxiety scores in the Elevated range at age 9.5-10 than at any other time point. When all children in the sample were considered, trajectories of parent-rated Generalized, Separation Anxiety, Panic and Agoraphobia symptoms overall remained relatively flat, showing no significant change over time. Moreover, there was little variability in trajectories between children across all domains, except Separation Anxiety. These findings expand on previous studies that have suggested developmental changes in anxiety in ASD by using a longitudinal design, focusing on concentrated developmental periods and highlighting differences in trajectories of change between anxiety domains. Moreover, the current study expanded measurement specificity to capture specific anxiety
disorder subtypes, rather than measuring anxiety as a broad construct (Bitsika & Sharpley, 2015; Davis et al., 2011; Gotham et al., 2015; Vasa et al., 2013).

When considered in parallel to the trends of change in anxiety development in the general population, the current data indicate that different anxiety disorder symptoms also show variable periods of developmental sensitivity in ASD. Based on population data regarding the age of onset and 12-month prevalence rates for each different anxiety disorder, the incidence of separation anxiety disorder shows a clear decrease from childhood to adolescence and into adulthood, while generalized anxiety disorder and panic disorder rarely occurs prior to adolescence but shows a stable incidence across adulthood (APA, 2013; Kessler et al., 2005; Kessler et al., 2012). Agoraphobia has a stable incidence from early childhood onwards (Kessler et al., 2012). The current data suggest that in ASD, parent perceptions of Generalized Anxiety, Panic and Agoraphobia symptoms remained at a stable level during middle childhood. However, unlike what is expected from typically developing children, parent-perceived Separation Anxiety levels remained stable rather than decreasing during this developmental period. This stability may be explained by the positive skew in Separation Anxiety observed in this sample overall, as apparent from the distributions and mean levels of anxiety relative to potential maximum scores at each time point. It is also possible that the trajectory of separation anxiety differed from that of typically developing children due to the delay in development caused by intellectual disability for a substantial proportion of children in this sample.

Another aim of the current study was to examine individual and parent predictors of anxiety outcomes. Language ability was significantly predictive of both parent-rated Generalized Anxiety and Separation Anxiety across ages 7.5-11, with higher levels of
anxiety reported in children who demonstrated age-typical language abilities. This result is consistent with a number of studies that have found a positive relationship between cognitive ability, adaptive functioning or language ability and broad measures of anxiety (Hallett, Lecavalier, et al., 2013; Lecavalier, 2006; Mazurek & Kanne, 2010; Simonoff et al., 2008; Strang et al., 2011; Sukhodolsky et al., 2008; Weisbrot et al., 2005). Nonetheless, whether a higher prevalence of clinically elevated anxiety should be expected in children with ASD who are higher-functioning, remains unclear. Given that it is certainly clear that children across levels of cognitive functioning experience anxiety, this positive relationship may be a product of parent-ratings being the main method of measurement in this literature. For example, it is possible that expressions of anxiety more frequently reported in children with ASD and intellectual disability (e.g., problem behaviours; Moskowitz et al., 2013) are less likely to be attributed to anxiety by outside raters, or captured by anxiety rating scales. Thus, including physiological and observational anxiety rating tools will be important in further clarifying this research question, particularly for children with both ASD and intellectual disability in future studies. Moreover, it is unclear whether a positive relationship would be apparent for language ability and other anxiety disorder domains. Therefore, future studies should also include anxiety measures specific to various anxiety disorder subtypes.

Furthermore, the positive relationship between language ability and anxiety level was graduated in the case of parent-rated Generalized Anxiety, but not for Separation Anxiety. It is likely that this finer difference is due to the verbal ability requirements implied by the items across the two subscales, similar to the findings of Hallett, Lecavalier, et al. (2013). Specifically, five of the six Generalized Anxiety subscale items ask parents to rate whether their child “worries” or “complains” about somatic symptoms or fears, whereas only two of
the six Separation Anxiety subscale items ask about “worries”. This result lends further support to the hypothesis that language ability likely moderates the expression of some types of anxiety, particularly as measured by rating scales completed by outside observers.

No sex differences in symptom levels of parent-rated Generalized or Separation Anxiety symptoms were apparent, consistent with several past reports on similar levels of anxiety between males and females (Gotham et al., 2013; Gotham et al., 2015; Park et al., 2012; Simonoff et al., 2008; Solomon et al., 2011). Notably, one theoretical commentary has put forward that the lack of sex differences in internalizing symptoms reported in the literature may be due to a bias in assessment tools and diagnostic practices (Kreiser & White, 2014). Kreiser and White (2014) argue that most comparisons across boys and girls have relied on brief parent rating scales (though see Oswald et al. 2016 for exception), thereby missing perspectives from the individuals themselves, other informants, or other measurement approaches. Future studies with large samples of female participants, relying on multiple informants and methods validated in ASD, will be needed to further clarify this pattern.

With regard to individual predictors, anxiety symptoms in children observed by parents as early as 1.5 years of age, positively predicted the tendency towards parent-rated Generalized and Separation Anxiety levels across middle childhood. These results suggest that parents can identify behavioural markers of future elevated anxiety early on in a child’s development. This has significant implications for early assessment practices as well as treatment planning. Querying for the presence of anxiety in early comprehensive autism diagnosis referrals will allow clinicians to anticipate the need for anxiety treatment. Incorporating brief parent rating scales into early ASD assessment practices is a cost and
time-efficient method of assessment (Sattler, 2002) that could serve this purpose. Further research will be needed to determine whether broad anxiety symptoms in early childhood are similarly predictive of elevated symptoms in other anxiety domains in ASD. Additionally, it will be important to determine whether direct observations of anxiety symptoms by clinicians early on in a child’s development are similarly predictive to parent-ratings.

Although the current study suggests that early assessment of ASD can potentially anticipate elevated anxiety symptoms in middle childhood, appropriate and effective approaches for preventing anxiety disorder development during the preschool years remain to be developed. Outside of a case study with a four year-old child (Nadeau, Arnold, Selles, Storch & Lewin, 2015), no children younger than seven years of age have been included in published studies of either individual-, group- or family-based delivery of cognitive-behavioural therapy for anxiety adapted for ASD (e.g., Reaven, Blakeley-Smith, Culhane-Shelburne, & Hepburn, 2012; Storch, Arnold, & Lewin, 2013; Wood et al., 2009). Given the level of verbal and cognitive abilities required to comprehend and apply the ‘cognitive’ components of cognitive-behavioural therapy, continuing to adapt existing treatments to be both developmentally appropriate and effective for children with ASD in the preschool years, is a task for future research.

The published literature on appropriate treatments for lower-functioning children is also in its infancy, possibly also due to the heavier emphasis on the cognitive aspect of established anxiety treatments (Hagopian & Jennett, 2008; 2014; Kreslins, Robertson & Melville, 2015). Using the behavioural approaches of applied behaviour analysis and positive behaviour support, a number of small studies - with samples ranging from one to three children - have treated fearful or avoidant responses in children with ASD and
intellectual disability (see Moskowitz et al., 2017 for review). In order to inform treatment planning and preventative efforts, more research on effective treatments for anxiety disorders in children with intellectual disabilities will be needed (Hagopian & Jennett, 2014).

Parental internalizing symptoms at the time of child’s diagnosis (ages 1.5-5 years) were predictive of the tendency towards parent-rated Generalized but not Separation Anxiety symptoms in middle childhood. Past studies have reported elevated symptoms of internalizing disorders in parents and close relatives of children with ASD (Hallett, Ronald, et al., 2013; Kuusikko-Gauufin et al., 2012; Micali et al., 2014), suggesting that anxiety and ASD may have shared hereditary components. The current study builds on these findings, suggesting this familial link may be deeper in the case of generalized anxiety, but not have a further connection with separation anxiety symptoms. The different pattern between the Generalized and Separation Anxiety outcomes suggests that the different anxiety disorders may be variable in their sensitivity to familial transmission. However, whether generalized anxiety symptoms are susceptible to familial transmission due to hereditary or parenting factors is not clear from these data. Considering that children with ASD are often unable to acquire skills through imitation due to the inherent social learning impairments of the disorder (Williams, Whiten, & Singh, 2004), it is possible they are protected from the environmental influence of elevated parental anxiety. Specifically, many skills that typically-developing children may learn naturally, in part by watching the social modeling of parents and peers, require explicit teaching for children with ASD (Williams et al., 2004). However, as children with ASD are less likely to emulate others’ behaviours – including their parents’ fearful, anxious or avoidant behaviours – their social learning impairment may serve as a buffer against the development of significant anxiety. Should the influence of
parental internalizing symptoms on the development of later generalized anxiety symptoms be evident in future studies, it will be important to parse out the roles of parental modeling, parenting behaviours and genetic influences. A clearer understanding of the etiology of anxiety disorders in ASD will also allow for consequent adaptation of intervention approaches.

Research and Clinical Implications

Results of Study 1 indicate that the SCAS-P, in its original format, is not a psychometrically sound tool for the assessment of anxiety in children with ASD, ages 7 to 11. This is a limitation of previously published research that used the SCAS-P as an outcome measure. However, two of the original subscales and the separated Panic and Agoraphobia Subscale show promise as parent-rated screeners of Generalized Anxiety, Separation Anxiety, Agoraphobia and Panic symptoms. Future research should evaluate the validity and reliability of these subscales with an ASD sample expanded to include older children. Psychometric evaluation of the complementary child-report version of the SCAS should also be conducted with an ASD sample.

Results of Study 2 shed light on trajectories of parent-perceptions of anxiety development in ASD during middle childhood. Given the peak in the proportion of children with “Elevated” Generalized and Separation Anxiety scores at age 9.5, future research should examine a possible correlation to developmental changes or common stressors around this age. Results of Study 2 also showed that, across the current sample, parent-rated Generalized Anxiety, Separation Anxiety symptoms, Panic symptoms and Agoraphobia symptoms remained stable over this time period. Moreover, there was little variance in trajectories across all but Separation Anxiety symptom domains. Additionally, several interesting findings emerged regarding predictive factors for anxiety development. Unlike girls in the
general population, girls with ASD were not at an increased risk for developing parent-reported Generalized or Separation anxiety symptoms in middle childhood. Parent ratings of anxiety in early childhood were significantly predictive of Generalized and Separation anxiety symptoms later on, suggesting that the risk for anxiety comorbid to ASD can be pre-empted as part of comprehensive assessments early on in the child’s development, and considered as a treatment target. Parental self-reported internalizing symptoms early on in their child’s life were predictive of increased Generalized Anxiety symptoms during middle childhood, though not of Separation anxiety symptoms. Assessment of parent’s symptoms early on in the child’s life can serve as a proxy for the symptom levels parents experienced prior to experiencing added stress and potential negative psychological consequences associated with the responsibilities of caring for a child with ASD. While assessments in the current study were conducted at the time of the child’s diagnosis and thus could not remove the considerable parenting stress that could result from this event (Keen, Couzens, Muspratt, & Rodger, 2010), this is an improvement on cross-sectional assessment. Thus, while these results may suggest that some anxiety disorders in ASD are not susceptible to the genetic impact of parental internalizing, further research on the relationship between parent and child psychopathology that includes data on parental psychopathology prior to their child’s birth, or ASD diagnosis either retrospectively (e.g., Micali et al., 2004) or as part of longitudinal designs would provide stronger evidence for this hypothesis.

Limitations

An overarching limitation of this dissertation was the scope of anxiety outcome measures. The anxiety measures used relied on parents’ perceptions and ratings, and were not informed by children’s own reports of anxiety, or by the perspectives of teachers or
clinicians. Given the robust evidence of discrepancies between informants across many childhood psychopathologies (De Los Reyes & Kazdin, 2005), multiple informant assessment can provide rich data on a child’s behavioural and emotional functioning. Although it has been suggested that individuals with ASD and high anxiety may have impaired insight into their own affective symptoms (White, Schry, & Maddox, 2012) and may have difficulty clearly articulating anxious thoughts (Ozsivadjian et al., 2012), self-report, clinician and teacher ratings can provide a fuller picture of a child’s functioning. Similarly, as anxiety is expressed cognitively, behaviourally and somatically, multi-method assessment that included physiological and observational measures would also have offered multiple converging sources of evidence. This limited measurement approach also meant that participants were not clinically assessed for comorbid psychiatric conditions, which could have allowed for a comparison between groups with and without comorbid anxiety disorders.

A second limitation of both studies was the lack of data on whether participating children were receiving either medical or psychosocial intervention for anxiety. There are emerging treatments shown to be effective for treating anxiety disorders in ASD, particularly in children without co-occurring intellectual disability (Kreslins, Robertson & Melville, 2015), and it is unknown whether the variance in rates of anxiety in the current sample was due to children receiving such treatments or other factors. Treatment could potentially alter the presentation and course of anxiety in children with ASD, and future studies should consider grouping children based on treatment status.

A further limitation of the research was the sample characteristics, which are somewhat different than the characteristics of epidemiological samples and thus limit the
generalizability of the findings. First, the sample includes children that were diagnosed up to age 5, although it is widely reported that children with milder autism traits are often not identified until later in childhood (Baio, 2014), when the social demands of their surroundings increase. Second, the parents responding were largely highly educated mothers, with 89.5 having received a post-secondary education. This rate is higher than the 64% post-secondary education rate reported across Canada (Statistics Canada, 2016), and of course, does not capture the perspectives or experiences of fathers. Finally, children with known genetic abnormalities were excluded from the sample. As different genetic disorders may also impact the presentation of anxiety, future research would benefit from examining how anxiety symptoms may present differently in children with different genetic disorders. Taken together, future research should aim to incorporate children with milder autism traits, as well as fathers, and parents with a representative distribution of education, in order to draw more generalized conclusions from this work.

Both studies also had unique limitations. A limitation of Study 1 was the treatment of items as continuous rather than ordinal for the overall CFA. This should be addressed in future studies with larger sample sizes. Moreover, although the SCAS-P was intended for 6 to 18 year-olds, the current data came from children 7 to 11 years of age: the findings may not generalize to older children. Study 1 was also limited by the potential challenges of responding to the SCAS-P for parents of non-verbal children. Parental response patterns have been reported to differ between parents of verbal and non-verbal children, with parents of verbal children endorsing more frequent presentations of verbally-loaded items on anxiety rating scales (Hallett, Lecavalier, et al., 2013). Hallett, Lecavalier et al., (2013) noted that such items tended to refer to the cognitive aspects of anxiety, such as “difficulty controlling
worries”, or verbal recognition of somatic or anxiety symptoms “complains about feeling sick when expects separation”. Thus, although it is clear that non-verbal children experience significant anxiety, items referring to verbally mediated anxiety within the SCAS-P were likely biased towards capturing the anxiety experiences of verbal children. While excluding non-verbal children, or those with IQ < 70 from this sample – as has been done in numerous other studies of anxiety measurement - could have potentially allowed for a clearer interpretation of the findings, this was outweighed by the benefit of including the often overlooked experiences of nonverbal children in this research.

With respect to the four SCAS-P subscales used as outcome measures in Study 2, it is important to note that research on the exact presentation of anxiety in ASD is in its early stages (Kerns et al., 2016; Rodgers et al., 2016). Accurately capturing and measuring the presentation of anxiety in ASD is an important challenge. As this area of research reaches consensus, the current findings on developmental trajectories and predictors of anxiety may need to be studied in ASD-specific anxiety domains. Moreover, due to the inappropriateness of the Social and Specific Phobia subscales to the current sample demonstrated in Study 1, the incidence of social anxiety and specific phobias across middle childhood was not assessed. Future research will be needed to determine whether these symptom trajectories follow a similar pattern. Particularly since cross-sectional studies in children and adolescents suggest Social Phobia and Specific Phobias to be some of the most frequently occurring anxiety disorders in ASD (van Steensel et al., 2011), it is possible that these symptom domains show differential patterns of growth during this developmental period.

A notable challenge in Study 2 was the subgrouping of children based on language ability. As no one measure that can capture the wide range of language functioning was
available, both the PLS-4 and the CELF-4 were used. Although both tools are psychometrically sound for the assessment of language abilities in ASD, the change in instrument limited the concordance of ability groupings. Unfortunately, the use of multiple measures to capture one widely ranging construct is a common occurrence in studies of cognitive and language development in children with autism (e.g., Bennett et al., 2014). Moreover, 12% of children did not have a language ability measure at age 8.5-9. As a consequence, language ability groupings further suffered from a lack of concordance as these participants were grouped based on cognitive ability results, or ADOS module selection at age 6.

Another limitation of Study 2 was the lack of a typically-developing control group. Having a direct comparison for the development of anxiety and its predictors using the exact same time points and measures as were used within this study could have allowed for a clearer understanding of the similarities and discrepancies with the general population. Clear comparisons with a typically-developing control could have also made the clinical implications of this study easier to apply for clinicians knowledgeable about anxiety disorders outside of ASD.

A further limitation of Study 2 was missing data in the case of the outcome measures, particularly at later time points, due to data collection being ongoing as part of an accelerated longitudinal design. More data was available at earlier ages (7 and 8) within middle childhood than later ages (9 and 10). Although the analytic approach was selected because of its robustness in cases with missing data, this research should be replicated with complete samples. Moreover, future longitudinal studies will allow for extension of this research into
adolescence and early adulthood, which are also developmental risk periods for anxiety disorders in the general population (Kessler et al., 2005).

**Strengths and Contributions**

The primary contribution of this dissertation was the examination of anxiety trajectories in middle childhood and early predictors based on longitudinal data in Study 2. This is the first known study to report on developmental changes in parent-rated Generalized Anxiety, Separation Anxiety, Panic and Agoraphobia symptoms in ASD based on longitudinal data. A strength within the design was the close evaluation of anxiety development through annual assessment during a critical developmental period in childhood. Each assessment was conducted during consistent and relatively brief developmental periods for all participating children. This allowed for a close focus on middle childhood, and overcame a limitation of the two other known longitudinal study of anxiety development in ASD (Gotham et al., 2015; Teh et al., 2017), which included multiple assessments over time but using an inconsistent assessment schedule over a very large age range (i.e., 6 - 24 years; 6-17 years). Furthermore, the results of Study 2 build on previous cross-sectional studies of factors that relate to anxiety in ASD through an evaluation of early predictors. This area of research sheds light on the mechanisms of development of anxiety in ASD, and is essential for the early identification of children with ASD at risk for developing anxiety disorders, for identifying an early developmental period for prevention efforts.

The results of Study 1 contributed to the growing literature that is evaluating validity of existing anxiety ratings scales in ASD samples (Lecavalier et al., 2014; Wigham & McConachie, 2014). It is the first study to examine the internal structure of the SCAS-P in an ASD sample ranging in cognitive ability. Moreover, as previous structural validation of
the SCAS-P in ASD had not suggested a replicable factor structure (Magiati et al., 2016), Study 1 provided the foundation for Study 2 in its suggestion of four psychometrically sound SCAS-P subscales. These four subscales are better suited for assessing parental perceptions of Generalized Anxiety, Separation Anxiety, Agoraphobia and Panic symptoms in children with ASD. Given the strong need to better understand and effectively treat anxiety in this population, psychometrically sound assessment tools are critical for future research efforts. These four subscales can inform the future adaptation of scales specific to ASD and serve as brief parent-report scales. Moreover, they can add new perspective to published studies that relied on the SCAS-P’s total or original subscale scores as outcome measures.

A strength of both studies was the inclusion of children across a range of cognitive functioning. The majority of studies that have reported anxiety rates or evaluated anxiety assessment methods in ASD have limited their samples to individuals with average to above average functioning (van Steensel et al., 2011). This has, in turn, limited our understanding about the presentation of anxiety and sound assessment practices for lower-functioning individuals. A final strength of both studies was that they examined a well-characterized sample of children (e.g., Bennett et al., 2014; Georgiades et al., 2012) who will continue to be followed until 19 years of age.

Conclusions

Research on the prevalence of anxiety in ASD (van Steensel et al., 2011), and its negative consequences (van Steensel et al., 2012) has preceded the development of solid psychometric tools for its measurement. A pressing need for research in this area resulted in studies reliant on measurement tools not validated in the ASD population (Lecavalier et al., 2014). Study 1 joins several reports on other existing anxiety measurement tools (Hallett, Lecavalier, et al., 2013; Stern et al., 2014; White et al., 2014) that indicate assuming the
validity of existing anxiety rating scales in this population is premature. However, adapted scale structures – such as that identified in this study – show promise. Moreover, longitudinal studies of anxiety development in this population can greatly inform our understanding and Study 2 offered rich information about anxiety’s development and early predictors. Further work is needed to expand this work to larger samples including older children with ASD and to include multiple-informants.
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Appendix A

Table 1

*Number and Proportion of Children whose Scores Fell within the “Elevated” Range on the SCAS-P*

<table>
<thead>
<tr>
<th>Age group</th>
<th>Score</th>
<th>7.5-8</th>
<th>8.5-9</th>
<th>9.5-10</th>
<th>10.5-11</th>
</tr>
</thead>
<tbody>
<tr>
<td>M</td>
<td>F</td>
<td>Total</td>
<td>M</td>
<td>F</td>
<td>Total</td>
</tr>
<tr>
<td>GAD</td>
<td>19</td>
<td>5</td>
<td>13.6%</td>
<td>23</td>
<td>7</td>
</tr>
<tr>
<td>SEP</td>
<td>21</td>
<td>8</td>
<td>16.8%</td>
<td>29</td>
<td>9</td>
</tr>
</tbody>
</table>

*Note.* GAD=Generalized Anxiety Subscale; SEP=Separation Anxiety Subscale; Total refers to the proportion of the sample (males and females) who exceeded the clinical cut-off.
Appendix B

Table 2

Parameter Estimates and Significance Tests for Unconditional and Unconditional Growth Models of Generalized Anxiety Outcomes

<table>
<thead>
<tr>
<th>Fixed effects</th>
<th>Unconditional Model</th>
<th>Unconditional Growth Model</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Coefficient (SE), t-ratio (df)</td>
<td>Coefficient (SE), t-ratio (df)</td>
</tr>
<tr>
<td>Intercept, $\beta_{00}$</td>
<td>2.44 (0.12) $t(234.35)=19.83^{***}$</td>
<td>2.32 (0.16) $t(204.70)=14.41^{***}$</td>
</tr>
<tr>
<td>Age Slope, $\beta_{10}$</td>
<td>0.06 (0.06) $t(170.03)=1.06$</td>
<td></td>
</tr>
<tr>
<td>Random effects</td>
<td>Variance (SD)</td>
<td>Variance (SD)</td>
</tr>
<tr>
<td>Intercept variance ($\tau_{00}$)</td>
<td>2.80 (1.67)</td>
<td>3.28 (1.81)</td>
</tr>
<tr>
<td>Residual ($\sigma^2$)</td>
<td>1.76 (1.33)</td>
<td>1.34 (1.16)</td>
</tr>
<tr>
<td>Age Slope, $r_1$</td>
<td>0.25 (0.50)</td>
<td>0.71</td>
</tr>
<tr>
<td>ICC</td>
<td>0.61</td>
<td></td>
</tr>
<tr>
<td>Deviance (-2LL)</td>
<td>2584.5</td>
<td>2562.1</td>
</tr>
<tr>
<td>Pseudo $R^2$</td>
<td>1-(1.34/1.76)=0.24</td>
<td></td>
</tr>
</tbody>
</table>

*Note.* ICC = Intraclass correlation coefficient; *** $p < 0.001$, ** $p < 0.01$, * $p < 0.05$
### Appendix C

#### Table 3

*Parameter Estimates and Significance Tests for Unconditional and Unconditional Models of Separation Anxiety Outcomes*

<table>
<thead>
<tr>
<th>Fixed effects</th>
<th>Unconditional Model</th>
<th>Unconditional Growth Model</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Coefficient (SE), <em>t</em>-ratio (<em>df</em>)</td>
<td>Coefficient (SE), <em>t</em>-ratio (<em>df</em>)</td>
</tr>
<tr>
<td>Interception, $\beta_0$</td>
<td>3.31 (0.18) <em>t</em>(231.55)=18.23***</td>
<td>3.46 (0.24) <em>t</em>(200.67)=14.69***</td>
</tr>
<tr>
<td>Age Slope, $\beta_{10}$</td>
<td>-0.07 (0.08) <em>t</em>(174.33)=-0.88</td>
<td>-</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Random effects</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Intercept variance ($\tau_{00}$)</td>
<td>6.36 (2.52)</td>
</tr>
<tr>
<td>Residual ($\sigma^2$)</td>
<td>3.17 (1.78)</td>
</tr>
<tr>
<td>Age Slope, $r_1$</td>
<td>0.45 (.67)</td>
</tr>
<tr>
<td>ICC</td>
<td>0.67</td>
</tr>
<tr>
<td>Deviance (-2LL)</td>
<td>2998.0</td>
</tr>
<tr>
<td>Pseudo $R^2$</td>
<td>1-(2.38/3.17)=0.25</td>
</tr>
</tbody>
</table>

*Note.* ICC = Intraclass correlation coefficient; *** $p < 0.001$, ** $p < 0.01$, * $p < 0.05$
### Appendix D

Table 4

*Parameter Estimates and Significance Tests for Unconditional, Unconditional Growth and Conditional Growth Models of Agoraphobia Anxiety Outcomes*

<table>
<thead>
<tr>
<th>Fixed effects</th>
<th>Unconditional Model</th>
<th>Unconditional Growth Model</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Coefficient (SE), t-ratio (df)</td>
<td>Coefficient (SE), t-ratio (df)</td>
</tr>
<tr>
<td>Intercept, $\beta_{00}$</td>
<td>0.74 (0.07) $t(233.06)= 10.27^{***}$</td>
<td>0.77(0.09) $t(250.60)=8.30^{***}$</td>
</tr>
<tr>
<td>Age Slope, $\beta_{10}$</td>
<td>-0.02 (0.03) $t(446.50)=-0.67$</td>
<td></td>
</tr>
<tr>
<td>Random effects</td>
<td>Variance (SD)</td>
<td>Variance (SD)</td>
</tr>
<tr>
<td>Intercept variance ($\tau_{00}$)</td>
<td>1.00 (1.00)</td>
<td>1.14 (1.07)</td>
</tr>
<tr>
<td>Residual ($\sigma^2$)</td>
<td>0.48 (0.69)</td>
<td>0.48 (0.69)</td>
</tr>
<tr>
<td>Age Slope, $r_1$</td>
<td>0.00 (0.04)</td>
<td>0.00 (0.04)</td>
</tr>
<tr>
<td>ICC</td>
<td>0.68</td>
<td>0.70</td>
</tr>
<tr>
<td>Deviance (-2LL)</td>
<td>1791.0</td>
<td>1788.8</td>
</tr>
<tr>
<td>Pseudo $R^2$</td>
<td>1-(0.48/0.48)=0</td>
<td></td>
</tr>
</tbody>
</table>

*Note. ICC = Intraclass correlation coefficient; *** $p < 0.001$, ** $p < 0.01$, * $p < 0.05$*
Appendix E

Table 5

Parameter Estimates and Significance Tests for Unconditional, Unconditional Growth and Conditional Growth Models of Panic Outcomes

<table>
<thead>
<tr>
<th></th>
<th>Fixed effects</th>
<th>Unconditional Model</th>
<th>Coefficient (SE), t-ratio (df)</th>
<th>Unconditional Growth Model</th>
<th>Coefficient (SE), t-ratio (df)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Intercept, $\beta_{00}$</td>
<td></td>
<td>0.38 (0.04)</td>
<td>t(235.96)=8.92***</td>
<td>0.38 (0.68)</td>
<td>t(191.1)=5.55***</td>
</tr>
<tr>
<td>Age Slope, $\beta_{10}$</td>
<td></td>
<td>0.001 (0.03)</td>
<td>t(158.8)=0.03</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Random effects</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Intercept variance ($\tau_{00}$)</td>
<td></td>
<td>0.25 (0.50)</td>
<td></td>
<td>0.35 (0.59)</td>
<td></td>
</tr>
<tr>
<td>Residual ($\sigma^2$)</td>
<td></td>
<td>0.44 (0.66)</td>
<td></td>
<td>0.40 (0.63)</td>
<td></td>
</tr>
<tr>
<td>Age Slope, $r_1$</td>
<td></td>
<td>0.02 (0.15)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>ICC</td>
<td></td>
<td>0.36</td>
<td></td>
<td>0.47</td>
<td></td>
</tr>
<tr>
<td>Deviance (-2LL)</td>
<td></td>
<td>1502.7</td>
<td></td>
<td>1500.5</td>
<td></td>
</tr>
<tr>
<td>Likelihood ratio test</td>
<td></td>
<td></td>
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<td></td>
<td></td>
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<tr>
<td>Pseudo $R^2$</td>
<td></td>
<td></td>
<td></td>
<td>1-(0.40/0.44)=0.09</td>
<td></td>
</tr>
</tbody>
</table>

Note. ICC = Intraclass correlation coefficient; *** $p < 0.001$, ** $p < 0.01$, * $p < 0.05$