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Repetitive Behaviours and Cognitive Flexibility in Children with High-Functioning Autism Spectrum Disorders

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Repetitive Behaviours and Cognitive Flexibility in Children with High-Functioning Autism Spectrum Disorders

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Abstract

This study examined the executive dysfunction hypothesis in children with high-functioning autism spectrum disorders (ASDs). This hypothesis involves a deficit in executive functioning, and specifically suggests that the repetitive behaviours in ASDs may stem from impairment in cognitive flexibility. Participants included 21 children aged 8-12 with high-functioning ASDs. Participants demonstrated impairment in a task of perseverative responding, a type of cognitive flexibility. However, in contrast to previous studies, this sample did not show significant levels of stuck-in-set perseveration in comparison to previously normed populations. Moreover, and in contrast to adults with ASDs, no correlations were found between degree of higher-order repetitive behaviours and cognitive flexibility, and lower-order motor repetitive behaviours and simple perseverative tasks. These results have implications for the developmental understanding of executive functioning and future investigations of the relationship between executive functions and behavioural symptoms of ASD.

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To my family, my warm love and many thanks.

Dedication

To my dad and the Rocky Mountains I wish he could have seen with me.

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List of Abbreviations

Abbreviation	Definition
AD	Autistic disorder
ADHD	Attention-deficit hyperactivity disorder
ADOS	Autism Diagnostic Observation Schedule
ADI-R	Autism Diagnostic Interview-Revised
AS	Asperger syndrome
ASD	Autism Spectrum Disorder
CF	Cognitive flexibility
D-KEFS	Delis Kaplan Executive Function Scales
DSM	Diagnostic and Statistical Manual of Mental Disorders
DSM-IV-TR	Diagnostic and Statistical Manual of Mental Disorders, 4th edition – Text Revision
EF	Executive functioning
fMRI	Functional magnetic resonance imaging
HFASD	High-functioning autism spectrum disorder
ICD-10	International Classification of Diseases, 10th edition
ID/ED shift	Intradimensional-extradimensional shift task
IQ	Intelligence quotient
MR	Mental retardation
PDD	Pervasive developmental disorder
PDD-NOS	Pervasive developmental disorder – not otherwise specified
RB	Repetitive behaviour
TMT	Trail Making Task
WASI	Wechsler Abbreviated Scale of Intelligence
WSCT	Wisconsin Card Sorting Test
YSII	Yale Special Interests Interview
YSIS	Yale Special Interests Survey

CHAPTER 1: INTRODUCTION

Autism Spectrum Disorders (ASDs) are a group of conditions that involve a qualitative impairment in social communication and stereotyped interests or behaviours (Pennington & Ozonoff, 1996). The spectrum is conceptualized as a continuum with varying degrees of severity of cognitive and adaptive ability, ranging from high to low levels of functioning. Individuals at the higher functioning end of the spectrum present with abnormal reciprocal social interaction skills in conjunction with fluent language skills, as well as stereotyped, repetitive behaviours (RBs), and average to superior intelligence (American Psychiatric Association, 2000). RBs include repetitive and/or circumscribed patterns of behaviour, interests, and activities that impair the ability of these individuals to form meaningful peer relationships (South, Ozonoff & McMahon 2005). In addition to these core impairments, a large body of research has found individuals with ASDs to be impaired in various areas of executive functioning (EF), providing support for the executive dysfunction theory of ASDs (Pennington & Ozonoff, 1994). Specifically, distinct impairment has been observed in the foundational executive function of cognitive flexibility (CF; Hill, 2004; Pennington & Ozonoff, 1996). Executive functions are cognitive processes that may serve as an intermediate link between brain bases of the disorder and behavioural symptoms (Gottesman & Gould, 2003). Support for this linkage has been found through studies of the relationship between CF and RBs in adults with ASDs (Lopez, Lincoln, Ozonoff, & Lai, 2005; South, Ozonoff, & McMahon, 2007). The present study aims to evaluate the developmental relevance of this research by extending this investigation to children with high functioning autism spectrum disorders (HFASDs).

Background and History

Autism was first mentioned over 60 years ago, and since that time research on this disorder has flourished. The term "autism" was initially used by Eugen Bleuler, a Swiss psychiatrist, in 1911 to define the disconnection to reality experienced in schizophrenia (Bleuler, 1911/1950 as cited in Klinger, Dawson, & Renner, 2003). In 1943, Leo Kanner described the core features of social impairment and stereotyped behaviours in relation to his observations of a group of 11 boys. Additionally, he observed that their language was abnormal: it was oftentimes developmentally delayed, and included episodes of echolalia, pronoun reversals, and mutism. In 1944 and independently of Kanner, Hans Asperger described a similar set of characteristics in a group of boys who displayed no language impairment. Despite having a large vocabulary and appropriate grammar use, this group had poor conversational abilities and social communication skills. Asperger's observations led to a distinctive diagnostic category for individuals who displayed social impairments and repetitive and restricted behaviours, but had no delay in developing spoken language.

Autism was first included in the North American diagnostic system: the Diagnostic and Statistical Manual of Mental Disorders (*DSM,* American Psychiatric Association, 1952) as Schizophrenic reaction, childhood type. Its reclassification as a neurobiologically-based disorder with a set of behavioural diagnostic criteria occurred in 1980 with the third revision of the *DSM* (American Psychiatric Association, 1987). As specific biomarkers have yet to be found for this group of conditions, ASDs continue to be diagnosed and defined according to behavioural and developmental criteria.

Overview of autism spectrum disorders. ASDs range in severity and include various subtypes but all share impairments in communication, social functioning, and RBs. ASDs are part of the broader group of pervasive developmental disorders (PDDs) that includes disorders wherein a developmental delay or regression in skill level occurs (American Psychiatric Association, 2000). The following are the current diagnostic criteria for ASDs according to the Diagnostic and Statistical Manual of Mental Disorders, 4th edition, Text Revision (*DSM-IV-TR*; American Psychiatric Association, 2000).

Autistic disorder. The diagnosis of Autistic Disorder (AD) requires the presence of evident impairment in social interaction, communication, as well as restricted and repetitive behaviours, interests, and activities. Delay in the individual's functioning must have been observed before the age of three years in social interaction, socially intended language or symbolic or imaginative play (American Psychiatric Association, 2000). Impairment in social interaction may include nonverbal impairments such as lack of eye contact, limited facial expressions, and gestures. Additionally, social interaction impairment may be evidenced through a failure to develop relationships with same-age peers, a lack of social or emotional reciprocity, and no spontaneous sharing of interests, enjoyment, or achievement with others (American Psychiatric Association, 2000). Communication impairment may be observed through a delay or absence of spoken language, inability to initiate or sustain conversation, idiosyncratic or repetitive use of language, and a lack of developmentally expected play (American Psychiatric Association, 2000). Restricted and repetitive behaviours may include stereotyped patterns of behaviour, motor mannerisms, ritualistic behaviour and inflexibility in routines, or an abnormal preoccupation with individual parts of objects (American Psychiatric Association, 2000).

The cognitive functioning levels of individuals with AD vary from significant cognitive disabilities to average or above average functioning. The descriptors *low-* (intelligence quotient (IQ) < 70) and *high-functioning* (IQ >70) are commonly used to characterize individuals based on cognitive functioning and adaptive ability (Tsatsanis, 2005). The cognitive functioning profiles of individuals with AD typically show strengths in visual-spatial processing and rote memory (Ghaziuddin & Mountain-Kimchi, 2004; Klin, Volkmar, Sparrow, Cicchetti, & Rourke, 1995; Lincoln, Allen, & Kilman, 1995; Mayes & Calhoun, 2003).

Asperger disorder. Currently, the DSM-IV-TR criteria for Asperger's Disorder, or Asperger syndrome (AS) as it is most commonly referred to in the literature, include qualitative impairment in: (a) social interaction, such as nonverbal impairments, failure to develop peer relationships, or a lack of spontaneous seeking to share enjoyment, interests, or accomplishments with others; and (b) restricted and repetitive patterns of behaviours, interests, and activities that are abnormal in their intensity or focus. The main diagnostic distinction of AS from AD is that these impairments may not be accompanied by any apparent delay in the development of language, cognitive ability, or developmentally appropriate self-help or adaptive skills (American Psychiatric Association, 2000).

The cognitive functioning profiles of individuals with AS often exhibit a strength in verbal reasoning abilities relative to their perceptual reasoning abilities (Ghaziuddin & Mountain-Kimchi, 2004; Lincoln, Courchesne, Allen, Hanson, & Ene, 1998; Ozonoff, South, & Miller, 2000).

Pervasive developmental disorder not otherwise specified. A diagnosis of PDD-NOS requires the presence of symptoms that are similar to those displayed in ASDs, but that do not meet the above criteria for AS or AD. This symptomatology may occur when symptoms develop later than suggested or when the symptoms or pattern of symptoms does not fit within the diagnostic criteria for other ASDs (American Psychiatric Association, 2000).

Other pervasive developmental disorders. Childhood Disintegrative Disorder and Rett's Disorder are two additional forms of PDD that occur with much less frequency than AD, AS, or PDD-NOS. Symptoms of Rett's Disorder appear after a period of typical normal prenatal and perinatal development, particularly normal psychomotor development throughout the first five months of life. These symptoms include: (a) a delay in the rate of head growth between the ages of five and 48 months of age; (b) loss of previously developed purposeful hand skills followed by the appearance of repetitive hand movements; (c) severe psychomotor retardation; (d) a loss of engagement in social interaction; and (e) the development of poorly coordinated gait or trunk movement (American Psychiatric Association, 2000). Childhood Disintegrative Disorder appears after an apparently normal course of development for the first two years of life. Symptoms manifest before the age of ten and are characterized by a loss of previously acquired skills in expressive or receptive language, social skills or adaptive behaviour, bowel or bladder control, play, or motor skills (American Psychiatric Association, 2000).

Prevalence. While a degree of uncertainty remains around the exact prevalence rate of ASDs (Fombonne, 2009), the most recent studies conducted in the United States estimate a prevalence rate of one affected child for every 88 children (Centre for Disease Control and Prevention, 2012). This class of disorder is more frequent in males, with an estimated occurrence of 1 in 54 males (Centre for Disease Control and Prevention, 2012). Prevalence is considered to be on the rise, with an increase of 78% observed in children the U.S. between 2002-2008 (Centre for Disease Control and Prevention, 2012). Since 2002, the largest increase in prevalence has been in cases of ASDs with borderline to above average cognitive functioning (IQ greater than 70), with a 93% increase seen in individuals with average to above average cognitive abilities (Centre for Disease Control and Prevention, 2012). The full cause for this dramatic increase is unclear, but increased awareness and identification methods account for at least part of the increase (Centre for Disease Control and Prevention, 2012). This dramatic surge in prevalence highlights the need for assessment, research, and intervention for this population.

Comorbidities. Comorbidity refers to the co-occurrence of two or more disorders in one individual. Given the substantial heterogeneity of symptom presentation in ASDs, the identification of disorders that are distinct from the core symptomology of ASDs is complicated (Sturmey & Sevin, 1994; Szatmari, Volkmar, & Walter, 1995). Nonetheless, several distinct patterns of comorbidity have been researched.

Intellectual functioning. Intellectual disability, classified as having an IQ score equal to or below 70, occurs in 38% of individuals with ASDs, with another 38% of the population demonstrating average or above average intellectual abilities, classified as having IQ scores of 85 and above (Centre for Disease Control and Prevention, 2012).

Mental health. Overall, anxiety disorders have a high rate of comorbidity with ASDs and have been reported to occur in approximately 40 to 84.1% of individuals (Bellini, 2004; Gillot, Furniss, & Walter, 2001; Gillot & Standen, 2007; Green et al., 2000; Kim, Szatmari, Bryons, Streiner, & Wilson, 2000; Muris, Steerneman, Merckelbach, Holdrinet, & Meesters, 1998; Simonoff et al., 2008) indicating that anxiety occurs more often in individuals with ASDs than in the general population (Bellini, 2004). Research on the nature of anxiety experienced by children with ASDs is in its infancy, but a higher prevalence of social phobia (Kim et al., 2000; Muris et al., 1998), specific anxiety disorders (Bellini, 2004; Gillott et al., 2001; Kim et al., 20000; Muris et al., 1998), generalized anxiety disorders (Bellini, 2004; Gillott et al., 2001; Muris et al., 1998; Russell & Sofronoff, 2005), and obsessive-compulsive disorder (Green, Gilchrist, Burton & Cox, 2000; Leyfer et al. 2006; Simonoff et al., 2008) have been noted.

Depression is estimated to occur in 2 -30% of individuals with ASDs, with higher estimates in children with AS (Ghaziuddin, Tsai, & Ghaziuddin, 1992; Ghaziuddin, Weidmer-Mikhail, & Ghaziuddin, 1998; Wing, 1981).

Attention-deficit hyperactivity disorder (ADHD) has been reported to occur in 14-78% of individuals with ASDs (Holtmann, Bolte, & Poustka, 2007; Keen & Ward, 2004; Lee & Ousley, 2006; Leyfer et al., 2006; Reiersen et al., 2007; Ruggieri, 2006; Simonoff et al., 2008; Sinzig, Walter, Doepfner, 2009; Yoshida & Uchiyama, 2004).

Medical comorbidities. Seizure disorders have been estimated to occur in between 7-14% of children with ASDs (Rapin, 1996; Tuchman, Rapin, & Shinnar, 1999) and in adults with ASDs between 20-35% (Minshew, Sweeney & Bauman, 1997). The prevalence of epilepsy is higher among individuals with ASDs who also have a cognitive disability (Amiet et al., 2008). The peak periods of risk for seizures are in early childhood and adolescence (Volkmar, & Nelson, 1990), and 30% of seizures occur in individuals with ASDs by 20 years of age (Rossi, Parmeggian, Bach, Santucci, & Visconti, 1995).

Sleep disorders are also common in children with ASDs, with prevalence rates ranging from 40-80% (Johnson, Giannotti, & Cortesi, 2009; Richdale, 1999) as compared to 30% in typically-developing children (Ferber, 1996).

Gastrointestinal disorders are estimated to occur in 9-70% of individuals with ASDs (Bauman, 2010). The range in estimates is due to the challenges surrounding the diagnosis of these disorders in the context of impaired language and communication skills as well as potentially dysregulated sensory processing (Bauman, 2010).

Sensory issues. Individuals with ASDs are often unusually sensitive to sensory stimuli (Liss, Saulnier, Fein & Kinsbourne, 2006). This symptom may manifest as an oversensitivity to certain stimuli that causes the individual to withdraw from it, or as undersensitivity to typically aversive stimuli such as bumps or bruises (Ornitz, 1988). Some individuals also exhibit an over-selective attention style characterized by exaggerated concentration on parts of objects or small details in the environment (Liss et al., 2006).

Outcome. Outcome for individuals with ASDs varies significantly. Many individuals exhibit significant impairment throughout their lives and reside with their families or in assisted living situations (Howlin, Goode, Hutton & Rutter, 2004), whereas others may find employment and live independently but continue to experience difficulties in forming and maintaining relationships (Green et al., 2000). The most predictive features of positive outcome in individuals with ASDs are the development of spoken language prior to the age of 5 and average to above average levels of cognitive functioning (Gillberg & Steffenburg, 1987; Howlin, Mawhood, & Rutter, 2000; Venter, Lord, & Schopler, 1992). Nonetheless, even individuals with high cognitive functioning may continue to have difficulties in regard to independent living, academic achievement, employment and social relationships (Green et al., 2000).

Executive Functioning

Theoretical models of the core deficits of ASDs play a key role in the search for their etiology and development. Several psychological theories have dominated research in this field, one of which has centered on a set of neuropsychological skills falling under the broader category of "EF" (Harris, 1993; Ozonoff, Pennington, & Rogers, 1991). EF is largely orchestrated by the prefrontal cortex, and is defined as "the ability to maintain an appropriate problem solving set for attainment of a future goal" (Welsh & Pennington, 1988 p. 201). More specifically, EF includes a number of interacting, yet theoretically distinct higher-order functions including inhibition, working memory, selective attention, planning, and cognitive and behavioural flexibility (Joseph & Tager-Flusburg, 2004). Collectively these functions are responsible for goal-directed behaviour through initiating and organizing mental resources, developing and initiating appropriate strategies for their achievement, inhibiting inappropriate actions, monitoring performance, and troubleshooting to enable an individual to carry out independent, self-serving, purposeful behaviour (Lezak, 1995). These functions are deemed executive because they form a “supervisory system” which processes information and integrates it along with previously stored knowledge (Shallice, 2004). Moreover, deficits in EF can impair various cognitive domains including learning, memory, language, and visual perception (Miyake, Friedman, Rettinger, Shah, & Hegarty, 2001). Although EF generally refers to higher-level skills, these functions influence skills and activities that range in complexity from abstract thinking and complex reasoning, to sustaining attention (Turner, 1997). Importantly, they are also involved in socio-emotional processes including modulating emotions, social self-awareness, and taking on other people’s perspectives (Anderson, Bechara, Damasio, Tranel, & Damasio, 1999; Anderson, Damasio, Tranel, & Damasio, 2000; Barrash, Tranel, & Anderson, 2000; Stuss & Alexander, 2000).

Foundational Executive Functions.

Inhibition. Inhibition is a foundational executive function that involves the ability to inhibit an initial, previously developed responding tendency, or the ability to put a stop to an already ongoing response (Barkley, 1997). Inhibition is essential for the development of self-regulatory functions such as deferring gratification and performing goal-directed and future-oriented actions (Barkley, 1997). It begins to develop in infancy and shows rapid improvement in early childhood (Best & Miller, 2010). By age four, children are able to perform simple and more complex inhibitory tasks (Best & Miller, 2010). More improvement occurs from ages 5-8, wherein children can perform tasks that tax both working memory and inhibition (Best & Miller, 2010).

Working memory. Working memory refers to the ability to maintain information in memory and manipulate it over brief time periods without the use of external cues (Alloway, Gathercole, & Pickering, 2006; Goldman-Rakic, 1987; Huizinga et al., 2006). Working memory improves significantly during the preschool years, and by age six a child is typically able to perform complex working memory tasks. Performance on such tasks improves until at least adolescence (Best & Miller, 2010).

Cognitive flexibility. CF is the executive function of interest to this study. It is defined as the ability to shift between rules or mental task sets and is considered to be an integral component of cognitive functioning (Gioia, Isquith, Guy, & Kenworthy, 2000; Husband & Miles, 1927; Lezak, 1995; Tinker, Imm, & Swanson, 1932). CF is continually in demand from the changes that occur in one's surrounding environment. It is in use when an individual is required to switch from one activity to another, to sequentially attend to various aspects of a problem for varying periods of time, or to participate in dynamic activities that have certain degrees of unpredictability. CF is a process that involves the integration of feedback from the environment as these dynamics shift, or rules and requirements change. An individual must monitor cues regarding changes, incorporate feedback, shift their attention, change mental set, and behave accordingly (Anderson, Damasio, Jones & Tranel, 1991; Gioia et al., 2000; Rothke, 1986; van der Sluis, de Jong, & van der Leij, 2004).

Although CF is considered a foundational executive function, it relies heavily on both working memory and inhibition. This is because prior to developing an ability to shift between mental sets a child must have the working memory capacity to remember a mental set and have an ability to inhibit the activation of an alternative set (Best & Miller, 2010).

Similarly to inhibition and working memory, CF improves with age (Best & Miller, 2010). Importantly, as CF depends on inhibition and working memory, it is less developed in preschool years (Best & Miller, 2010). Indeed, it has been shown that preschoolers are able to successfully shift between mental sets only when inhibitory demand is reduced (Rennie, Bull & Diamond, 2004) or when set rules are contextualized within a story (Hughes, 1998). Further improvements continue through to adolescence (Davidson, Amso, Anderson, & Diamond, 2006; Huizinga, Dolan & Van der Molen, 2006; Luciana & Nelson, 1998)

CF impairment can appear as either simple perseverative responding or stuck-in-set perseveration (Turner, 1999). Simple perseverative responding occurs wherein an individual cannot regulate their prepotent, or previously repeated and reinforced, response tendencies despite being asked to change their responding (Turner, 1999). Stuck-in-set perseveration occurs when an individual cannot shift their thinking between the different sets of rules required by different aspects of a task (Turner, 1999).

The Executive Dysfunction Hypothesis of Autism Spectrum Disorders

The Executive Dysfunction hypothesis (Hughes, Russell, & Robbins, 1994; Ozonoff et. al., 1991) attributes autistic symptomatology to impairments in EF. This theory holds appeal because it has the potential to explain some characteristics that are beyond the scope of other theories (Turner, 1997). Through this framework, researchers have attempted to examine the relationship between EF and language impairments (Joseph, McGrath, & Tager-Flusberg, 2005; Landa & Goldberg, 2005), social impairments (Landa & Goldberg, 2005) and RBs (Lopez, Lincoln, Ozonoff, & Lai, 2005; South, Ozonoff, & McMahon, 2007). Specifically, executive dysfunction has been proposed as an explanation for the RBs that span the autism spectrum (Turner, 1997, 1999). Linking cognitive processes to observable behavioural symptoms of a psychiatric disorder serves to establish an intermediate connection between these symptoms and the genetic or brain bases for the disorder (Gottesman & Gould, 2003). RBs may be the direct result of executive dysfunction, wherein the individual lacks the ability to typically generate, plan, and control behaviour (Ridley, 1994). This is a compelling explanation as it could account for the range and variety of RBs in ASDs, their pervasive nature, and the resistance to intervention that is described in studies of treatment response (Epstein, Doke, Sajwaj, Sorrel, & Rimmer, 1974; Harris & Wolchik, 1979). Moreover, the repetitive, rigid, and invariant nature of the behaviour could be explained by executive dysfunction as an inability to regulate behaviour effectively (Turner, 1999).

What are Repetitive/Stereotyped Behaviours?

Repetitive, stereotyped behaviours have been considered a prominent feature of ASDs since the earliest descriptions of their symptomatology (Asperger, 1944; Asperger, 1991; Kanner, 1943), and are included in the International Classification of Diseases, 10th Edition (World Health Organization, 1993) and *DSM-IV-TR* (American Psychiatric Association, 2000) diagnostic systems. However, this feature has historically been the most neglected symptom in research on ASDs (Baron-Cohen, 1989; Shao et al., 2003; Turner, 1999), and our understanding of the causality, maintenance, function, and appropriate intervention for these behaviours is limited (Turner, 1999).

RBs in ASDs are a broad class of behaviours characterized by rigidity, invariance, repetition, and inappropriateness (Turner, 1999). This category includes spontaneous dyskinesias, stereotyped motor movements, attachment to specific objects, repetitive object manipulation, repetitions in speech, and narrow, circumscribed interests (Turner, 1999). These behaviours can be divided into two categories: higher-level or complex behaviours such as attachment to specific objects, resistance to change, repetitive language or echolalia, and circumscribed interests; and lower-level behaviours often expressed as repetitive movements such as tics, dyskinesias, stereotyped movements, self-stimulatory behaviour and repetitive object manipulations (Turner, 1999). While all of these behaviours are unusual, stereotyped, and persistent, there is otherwise little agreement as to whether they make up a single symptom category (Turner, 1999).

While children with HFASDs display some lower-level behaviours (South, Ozonoff, & McMahon, 2005), they also present with higher-order RBs such as circumscribed interests (McPartland & Klin, 2006). Children with circumscribed interests often acquire a large amount of information about a very specific area of interest that may be atypical or developmentally inappropriate (McPartland & Klin, 2006). The topics of interest may differ from those of a typically-developing child's in their eccentricity, or inappropriateness for the individual's developmental level (McPartland & Klin, 2006). Other interests may be quite ordinary in their focus, but atypical in their intensity (McPartland & Klin, 2006). Furthermore, whereas typically-developing children may become very interested and knowledgeable about certain subjects, individuals with HFASDs are distinguished by their obsessive nature and depth of knowledge regarding the topic (Volkmar & Klin, 2000). Unlike the specific skills sometimes exhibited in AD that are more likely to involve visuo-spatial tasks, music, object manipulation or savant skills, the focus of circumscribed interests is on gathering an extraordinary amount of information about one topic often with a zeal that is far above that of a typically-developing child (Volkmar & Klin, 2000). Moreover, the topics of interest are often pursued without a broader understanding of their significance (McPartland & Klin, 2006). The specific topics of interest may change across development, but can take over the child's and/or family's life, and also interfere with social interactions (Volkmar & Klin, 2000). Although children with HFASDs may actively desire social relationships and interaction, they are often too intent on sharing their enthusiasm for their circumscribed interest with others (McPartland & Klin, 2006). This interest contributes to a behavioural manner that is often perceived as awkward or odd (McPartland & Klin, 2006).

The neglect of repetitive behaviour in research. Despite the significant impairment caused by circumscribed interests and other RBs seen in ASDs, as well as their inclusion as important features of AS and AD in Asperger's (1944) and Kanner's (1943) original descriptions, this core symptom has received comparably little attention in the research literature (Turner, 1997).

One reason for this neglect is that RBs are not specific to ASDs. Such behaviours often occur in typically-developing individuals, particularly in childhood (Thelen, 1979), or in conjunction with boredom or anxiety (Thelen, 1979, 1981). Moreover, some classes of RBs are also characteristic of other clinical conditions including schizophrenia, obsessive-compulsive disorder, dementia, Parkinson's disease, Tourette's syndrome, and mental retardation (MR) (Ridley, 1994; Turner, 1995). However, this non-specificity of RBs to ASDs does not minimize their significance to ASDs, as atypical language or social skills are also features of some of the conditions above (Turner, 1997).

Another reason for the neglect of RBs is that MR is present in a majority of individuals with ASDs, and the high comorbidity has incorrectly contributed to the idea that RBs are simply an indicator of intellectual impairment or of organic dysfunction (Turner, 1997). However, this idea is inconsistent with a number of findings. Firstly, some higher-level RBs such as circumscribed interests and an insistence on maintaining rigid and invariant routines are restricted to or are distinctly more prevalent in ASDs than other disorders (Frith, 1989; Lord & Pickles, 1996; Turner, 1999). Secondly, there is some evidence that individuals with ASDs exhibit significantly more RBs than control subjects matched for level of intellectual impairment (Turner, 1995). Lastly, individuals at the higher end of the autism spectrum, whose intelligence is in the average or superior range, nonetheless exhibit RBs (Bishop, Richler, & Lord, 2006; South et al., 2005; Szatmari, Bartolucci, & Bremner, 1989; Tantam, 1991, 2000). These findings are incompatible with the idea that RBs are a result of intellectual impairment, and indicate that they are a core symptom of ASDs (Turner, 1997, 1999).

Another assumption about RBs that has contributed to their neglect is that they serve to reduce anxiety, and are simply an adaptive coping strategy that is secondary to the disorder (Tantam, 2000; Turner, 1999). This assumption has stemmed from the commonly adopted theory to explain RBs: the hyperarousal theory first described by Hutt and Hutt (1965; 1970), and by Hutt, Hutt, Lee, and Ounsted (1964). This theory suggests that RBs are a regulatory mechanism for maintaining optimal sensory input, necessary as individuals with ASDs experience chronically high levels of arousal. Thus, it is suggested that RBs act to block sensory input from the environment. Moreover, the theory suggested that novel situations or stimuli are arousing and are thus met with avoidance; this results in the “insistence on sameness” that has been consistently observed across individuals with ASDs. The empirical basis for this theory, however, has also been questioned (Turner, 1997, 1999). Turner asserts that this account is based on circular reasoning: the observation of high arousal in autistic individuals stems from their RBs, and the behaviour is then explained as a response to this same high arousal. This is in part due to a lack of ascertainment that individuals with ASDs are more physiologically aroused than groups without ASDs (Rogers & Ozonoff, 2005), or that increases in environmental stimulation result in more RB (Turner, 1997, 1999). Indeed, the hyper-arousal account is inconsistent with the display of RBs across a variety of environmental conditions, and particularly in those where there is no apparent stimulation (e.g., time-out rooms).

Repetitive Behaviour as Stemming from Executive Dysfunction

The EF hypothesis provides an alternative avenue through which to understand RB (Lopez et al., 2005; Pennington, 2002; Turner, 1997, 1999). Exploring RB through the lens of executive dysfunction may aid in linking cognitive impairment to this key symptom (Turner, 1997, 1999). If executive dysfunction is related to the development of symptomatology in ASDs, it follows that this impairment be universal to all individuals with ASDs, that the impairment be specific to ASDs, and that the degree of impairment be correlated with the severity of symptomatology that is hypothesized to arise from this impairment (Turner, 1997). If this last prediction is true in regard to RBs, that is, the severity of EF deficit correlates with an increase in the expression of RB, the relation between EF and the symptom is very likely significant (Turner, 1997).

Turner (1997) has proposed a potential EF pathway for the maintenance of these behaviours in AD. She suggests that RBs are an extension of flawed processing in the frontal lobes and specifically, of the tendency to perseverate. The individual becomes “locked into” one set of behaviours, and a failure to inhibit the self-generated actions results in RBs. Impairment in the inhibition of thoughts, actions, and behaviours would cause the individual to rigidly continue the initiated behaviour. This account conceptualizes RBs as the behaviourally expressed counterpart of a tendency to cognitively perseverate.

Turner (1997) hypothesizes that impairments in CF as observed through laboratory-based measures should be highly correlated with the extent of RB displayed by the individual. More specifically, Turner predicts that CF as simple perseveration of a previous response will be correlated with low-level RB, such as stereotyped movements or manipulations of objects. In contrast, she predicts that CF as stuck-in set perseveration - that seen when an individual is unable to redirect their attention to a newly presented set of rules, but instead persists with the original set - will be correlated with higher-level RBs, such as circumscribed interests, repetitive language, and rigid routines and rituals. Turner's hypothesis is called the "failure of behavioural inhibition" hypothesis (Turner, 1997).

Cognitive flexibility impairment in autism spectrum disorders. In support of this seminal hypothesis, there is significant evidence that CF is impaired in individuals with ASDs (Hill, 2004; Pennington & Ozonoff, 1996). Inflexibility or impairment in CF is seen by both parents and clinicians as one of the most difficult to manage and persistent features apparent in ASDs (Geurts et al., 2009). It is evident when individuals have difficulty switching to new strategies in daily activities, or when they cannot shift between perspectives in their interactions with others (Geurts et al., 2009). In addition to their occurrence in daily functioning, these impairments have also been demonstrated using neuropsychological measures of CF (Manjiviona & Prior, 1999; Ozonoff et al., 1991; Ozonoff, Rogers, & Pennington, 1991; Wilcutt, Sonuga-Barke, Nigg, & Sergeant, 2008). Indeed, impairment in CF has been found in individuals with ASDs in comparison to IQ-matched clinical controls, including individuals with ADHD (Nyden, Gillberg, Hjelmquist, & Heiman, 1999; Ozonoff & Jensen, 1999), dyslexia (Bennetto, Pennington, & Rogers, 1996; Ozonoff & McEvoy, 1994) as well as to age-, gender- and IQ-matched typically-developing controls (Prior & Hoffmann, 1990; Rumsey & Hamburger, 1988, 1990).

Neuropsychological measures of cognitive flexibility. Neuropsychological measures of CF require examinees to alter their behaviour or thought process relative to the changing demands of a task (Hill, 2004). CF tasks typically rely on switching between two or more mental sets—with each set possibly containing several task rules (Crone, Somsen, Zanolie, & Van der Molen, 2006). One type of CF impairment is stuck-in-set perseveration: that observed when an individual has difficulty switching between mental sets. Stuck-in-set perseveration has most commonly been tested through the Wisconsin Card Sorting Task (WCST), a problem-solving task that asks examinees to view and sort cards in a deck with differing types, colours, and numbers of symbols on each card by placing them in front of four stimulus cards. After each placed card, the examinee is given feedback as to whether the placement is correct according to a principle that is unknown to the examinee, and that may change without warning. The examinee's working memory is activated in the maintenance and updating of each matching principle based on the continuous feedback (Best & Miller, 2010). Perseverative errors, made through continued responding based on the previous mental set, indicate stuck-in-set perseveration (Hill, 2004).

Performance on this task is often impaired in individuals with ASDs, as evidenced by significant perseveration in responding following a change in principle despite negative feedback (Ozonoff et al. 1991; Prior & Hoffman, 1990; Rumsey, 1985; Rumsey & Hamburger, 1988; Szatmari et al. 1990) though some studies have failed to find a difference between typically-developing and children with AS (Nyden et al., 1999) and adults (Minshew, Goldstein, Muenz & Payton, 1992). However, even when developmentally adjusting the task for preschool-aged children, McEvoy, Rogers and Pennington (1993) found that preschool-aged children with ASDs made significantly more perseverative errors than age-matched controls. Moreover, it has been found that children with ASDs and IQ in the average range demonstrate stable impairments on a neuropsychological measure of CF over a 2.5 year period (Ozonoff & McEvoy, 1994), indicating that perseveration is a lifetime feature of the disorder.

Empirical investigations. Despite significant evidence of impairment in CF in ASDs, only a few studies have examined the theoretically proposed link between RB and CF deficits in ASDs (Lopez et al., 2005; South et al., 2007). Lopez and co-authors (2005) examined the relationship between RBs and EF in a group of adults with AD (n=17), in comparison to a group of typically-developed adult controls (n=17). RBs were measured through a composite of scores derived from the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DiLavore, & Risi, 1999), the Autism Diagnostic Interview-Revised (ADI-R; Lord, Rutter, & Le Couteur, 1994), the Gilliam Autism Rating Scale (Gilliam, 1995), and the Aberrant Behaviour Checklist-Community (Aman & Singh, 1986) and EF was measured through a composite of the Delis Kaplan Executive Function Scales (D-KEFS; Delis, Kaplan & Kramer, 2001) and the WCST. The authors found that RBs were related to the EF processes of CF, working memory, and response inhibition, but not planning or fluency. In further analyses, CF was the only impaired executive function that uniquely predicted a significant portion of the variance in the composite measure of RB and it predicted more RB than performance or verbal measures of IQ. However, when entered into a regression model with working memory and response inhibition, no executive function variable predicted RBs. Rather, the findings suggested that RBs may be the result of an EF profile of strengths in some processes and deficits in others. A notable limitation of this study is the composite method used to measure RBs (Barber, 2008), a method that may reflect a general symptomatology of AD rather than RBs alone (Turner, 1997). Moreover, Turner's hypothesis would predict that the stuck-in-set perseveration found by the authors would have correlated primarily with higher-level RBs; however, higher-level RBs were not partitioned out in the analysis.

This limitation was avoided by South and co-authors (2007) who used specific measures of RBs in a study of their relationship to measures of weak central coherence and EF. Their sample included 19 individuals with HFASDs age and IQ-matched to 18 typically-developing controls, with a mean age of approximately 14 in both groups. RBs were measured with the Repetitive Behavior Interview (RBI; Turner, 1997) and the Yale Special Interests Interview (YSII; South, Klin, & Ozonoff, 1999). As the RBI does not extensively measure circumscribed interests, the YSII - a measure of circumscribed interests - was used as a complementary measure. Additionally, the RB domain scores from the ADI-R and ADOS were used. Significant correlations were found between perseverative responses on the WCST, and the RB domains of the ADOS and ADI-R, but not of the YSII or the RBI. The authors hypothesized that this result could have resulted from the composite nature of the ADOS and ADI-R scores, but found no significant correlation between perseverative scores and a combined score of the YSII and RBI with further analyses.

In addition to these examinations of the relationship between CF and RBs, one recent study has linked brain functioning during a task of CF to RBs (Shafritz, Dichter, Baranek & Belger, 2008). Eighteen individuals with high-functioning AD and 15 typically-developing controls performed a computer-administered task of CF while undergoing functional magnetic resonance imaging (fMRI). In the task, participants saw sequences of shapes and were asked to alter their responses by pressing on one of two buttons based on whether or not the shape on the screen was a preassigned target shape. Thus, non-target were incorporated into the sequence as distractors but also required a button-press. The participant was informed through a message on the screen when the assigned target shape changed several times throughout the task. This process resulted in condition one, wherein participants had to change their response solely based on the appearance of a target shape, and condition two, where participants had to change their response based on the appearance of a newly assigned target shape. The authors argued that errors in the first condition reflect simple perseverative responding, whereas errors in the second condition reflect stuck-in set perseveration. While this task was used to mimic features of the traditionally used WCST, it differed specifically in that it informed participants of the upcoming change in cognitive set rather than requiring participants to shift set based on negative feedback. Tasks in which rules are explicitly expressed are typically considered to tax inhibition rather than CF (Best & Miller, 2010). Moreover, the task had not been previously used with samples with ASDs. Indeed the authors found no impairment in individuals with ASDs in the second condition, possibly as participants were overtly informed of the change in set (Shafritz et al., 2008). Nonetheless, it was found that relative to the control group, the ASD group was impaired in condition one, exhibiting simple perseveration in responding regardless of whether there had been a change in the set of rules (Shafritz et al., 2008). Moreover, in the ASD group, the severity of RBs as measured by the ADI-R RB domain was negatively correlated with activation to target shapes in the left intraparietal sulcus and anterior cingulate regions. This finding links brain activation patterns during a task of CF with a core symptom of ASDs.

Together, the studies above suggest that CF may be an intermediate marker of both RBs and neurobiological dysfunction in ASDs.

Summary and Critique

There is a growing body of evidence relating impairment in EF to ASDs. In particular, there is consistent empirical support for an impairment in CF as demonstrated by perseverative errors on measures such as the switching section of the Trail-making Task (TMT; Lopez et al., 2005) and stuck-in set perseveration on the WCST (Bennetto et al., 1996; Nyden et al., 1999; Ozonoff & Jensen, 1999; Ozonoff et al., 1991; Ozonoff & McEvoy, 1994; Prior & Hoffman, 1990; Shu, Lung, Tien, & Chen, 2001; Szatmari, Tuff, Finlayson, & Bartolucci, 1990).

Several recent studies have examined the connection between impairment in EF and core, behavioural symptoms of ASDs (Lopez et al., 2005; South et al., 2007). These studies have found a link between the rigidity and inflexibility seen in RBs and CF in adults with ASDs, suggesting that the EF hypothesis may serve to explain the cognitive processes contributing to these behaviours. However, no study has yet extended this investigation to a younger age group with ASDs. Given the developmental nature of CF, evaluating the relationship between CF and RB in children of limited age ranges is key towards evaluating the EF hypothesis in this domain.

Furthermore, previous studies have evaluated a general connection between CF and severity of RB, rather than dissecting these behaviours (Lopez et al., 2005; South et al., 2007). In accordance with Turner's (1997) hypothesis, higher- and lower-order RBs will correlate to higher- and lower-order CF, specifically, stuck-in-set perseveration and simple perseverative responding. Thus, sectioning off both RBs and CF into component parts is important in evaluating Turner's hypothesis.

The Present Study

This investigation was conducted within the context of a larger research project *Resilience in Children with High-Functioning Autism Spectrum Disorders,* that was conducted through the University of Calgary. This larger study is looking at a number of factors including inhibition, working memory, emotional resilience, emotional intelligence, social skills, and socio-emotional well-being. Each of these factors is being evaluated individually. The overall goal of this research study was to evaluate factors that predict positive outcomes and strengths in this vulnerable population.

The present study looks at a subset of data collected through the larger study. It aimed to answer three research questions.

Research questions.

1. Do children with HFASDs exhibit impairment in CF? Specifically:
    a. Do children with HFASDs exhibit stuck-in-set perseveration on a task of set-shifting relative to the typically-developing population?
    b. Do children with HFASDs exhibit perseverative responding on a task of simple perseveration relative to the typically-developing population?
2. In children with HFASDs, does stuck-in-set perseveration correlate with time spent on higher-order RBs?
3. Does perseverative responding correlate with severity of lower-order RBs?

Hypotheses. Past research investigating EF in children and adults with ASDs was used to inform the following hypotheses:

1. Given that previous research has demonstrated impairment in CF in children with ASDs in comparison to the typically-developing population (Hill, 2004), it was hypothesized that study participants will likewise demonstrate such impairment in comparison to the normed population on two standardized measures of CF.

   a. It is expected that stuck-in-set will be evident through more perseverative errors on the WCST than expected based on the norming sample.
   b. It is expected that perseverative responding will be evident through a larger amount of time required to complete the TMT4 of the D-KEFS, relative to the normed population.

2. In accordance with Turner's (1997) "failure of behavioural inhibition" hypothesis, it is expected that a lack of CF as measured by degree of stuck-in-set perseveration will positively correlate with time spent on higher-order RBs.
3. Also in accordance with the "failure of behavioural inhibition" hypothesis (Turner, 1997), CF as measured through simple perseverative responses will positively correlate with the frequency and markedness of repetitive motor movements exhibited by the child.

CHAPTER 2. METHOD

Participants

Demographic and clinical characteristics of the participant group are summarized in Table 1. Participants were 21 children between the ages of 8-12 previously diagnosed with AD, AS, or PDD-NOS by a mental health professional licensed and trained to make such a diagnosis.

Twenty of the participants were male, and one participant was female. This discrepancy is to be expected given the distribution of the population diagnosed with ASDs, which has an estimated male to female ratio of 4.6:1 in ASDs overall and a higher ratio in individuals with average to above average cognitive functioning (Centre for Disease Control and Prevention, 2012). The average age of the participants was 10.2 years. Parents provided information about the child's official diagnosis and the specific ASDs by way of demographic questionnaire. Twenty-nine percent had a diagnosis of AD, 52% had a diagnosis of AS, and 19% had a diagnosis of PDD-NOS.

Participants completed the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999) at the outset of their testing session, and participants with scores lower than 85 on the Verbal or Performance IQ were not included in the remainder of the study so as to ensure that participants would understand task requirements and that they met criteria for the average to above-average cognitive functioning of HFASDs. The verbal IQ of participants ranged from 86-151 with a mean of 117.38 and standard deviation of 14.46. The performance IQ ranged from 96-147 with a mean of 119.86 and a standard deviation of 14.87.

Measures

Demographic information. A questionnaire was created to gather information from the parent regarding the participating child's gender, date of birth, grade, diagnoses, history of head injuries, and other relevant demographic information.

Diagnostic criteria. Participants were required to have a previous diagnosis of ASDs made by an appropriately-licensed professional prior to participating. Diagnoses of ASDs were confirmed through the ADI-R (Rutter, LeCouteur, & Lord, 2003).

Autism Diagnostic Interview-Revised. The ADI-R is a semi-structured, standardized interview administered by a trained interviewer to a parent or guardian of the individual suspected of having an ASD. It contains 93 questions that are grouped into background information, language and communication, social development and play, repetitive and restricted behaviours, and general behaviour concerns sections. The informant should be familiar with the individual's developmental history and current functioning. It is appropriate for individuals at a developmental level of 24 months and above. Answers are scored on a four-point scale according to the presence and severity of specific behaviours. These scores are inputted into a diagnostic algorithm that is divided into the following domains: Communication; Restricted, Repetitive and Stereotyped Patterns of Behaviour; Reciprocal Social Interaction; and Abnormality of Development at or Before 36 months. To be classified as meeting criteria for any of the ASDs, scores must reach specific cut-off points in each domain. For domain scores, inter-rater reliabilities ranged from .52-.97; test-retest reliabilities within 2-5 month time periods ranged from .93-.97. The validity of the ADI-R was determined on a sample of 226 children previously diagnosed with an ASD. Concurrent validity with independently formulated clinical diagnoses was very good (mean kappa = .74), and criterion validity with the prior version of the

ADI was excellent. Both individual questions and domain scores demonstrate good discriminative validity, with a sensitivity of 1.0 and a specificity greater than 0.97, indicating that the ADI-R is effective in differentiating between individuals with and without ASDs (Rutter et al., 2003).

Wechsler Abbreviated Scale of Intelligence. Participants were required to obtain a PIQ and VIQ of 85 or higher on the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999) to be included in the study. The WASI is an abbreviated measure of intelligence that requires 30-60 minutes for administration time. The VIQ is a measure of verbally-based knowledge and verbal reasoning. The PIQ is a measure of visual spatial reasoning, ability and logical thinking. Both the VIQ and PIQ scores are presented as standard scores (M=100, $SD=15$).

Internal reliability coefficients for children ages 6 to 16 are reported to be .93 for VIQ, and .94 for PIQ (Wechsler, 1999). The more comprehensive Wechsler Adult Intelligence Scale, Fourth Edition, has been compared to the WASI and a concurrent validity has been demonstrated through correlations of .92 (Axelrod, 2002). Similar patterns of correlation have been reported with a small sample of Canadian children (Saklofske, Caravan, & Schwartz, 2000).

Repetitive behaviours. Lower-level RBs observed by parents in either the participant's past and present daily functioning were assessed through the RB domain of the ADI-R. The RB domain of the ADI-R measures parental report of motor mannerisms exhibited by the child in either the hands or complex movements of other body parts, or the whole body. The RB domain provided a quantitative measure of severity and frequency of RBs that had been exhibited by the child currently or at any time in the past. This quantification is based on the interviewer's judgement of the parent's answer to the interview questions. This domain was used as it provided the desired specificity to measure the motor class of RBs.

Circumscribed interests, classified as higher-order RBs, were assessed through parent report on the Yale Special Interests Survey (YSIS; Volkmar & Klin, 1996). Specifically, the proportion of time the participating child has been occupied by their circumscribed interest throughout both their preschool and elementary-school aged years was the primary measure of interest.

The Yale Special Interests Survey. The YSIS (Volkmar& Klin, 1996) is a semi-structured parent-report questionnaire designed to gather information about an individual's circumscribed interests. This measure was selected due to its specificity to circumscribed interests. It asks about three developmental stages, two of which applied to the current sample: preschool and school-age. For example, for the preschool age period the questionnaire asks the reporting parent whether the child has had "an unusually intense interest at this age in one or two topics or activities, more so than other preschool-age children". It then probes for examples of the topic of the child's circumscribed interests, and asks about any unusual skills the child may have had. Lastly, it asks about the amount of child's time and the proportion of conversation with family, peers, and other adults that was taken up by the circumscribed interest. An identical set of questions is asked for the school-age period. Proportion of time is measured as a percentage of the child's time (Less than 25% of the time, 25-75% of the time, 75% or more of the time) and is then translated into a score of 1, 2 or 3. Primary analyses from the YSIS used a summary score of the amount of time taken up by circumscribed interests in both the preschool and school-age years. This resulted in a score ranging from 1 to 6 that represented a sum of time spent on circumscribed interests in their preschool- and elementary-school age years. No measures of validity are reported for the survey. Good test-retest reliability has been reported for the interview based on its administration to over 800 participants (Klin, 1999 as cited in South et al. 2005).

Cognitive Flexibility.

Delis-Kaplan Executive Functioning Scale. The D-KEFS (Delis et al., 2001) is a comprehensive measure of EF processes including inhibition, fluency, verbal abstraction, impulse control, concept formation, abstract thinking, planning, reasoning, and CF in both the verbal and spatial modalities. It contains nine subtests that may be administered individually as well as altogether. No composite scores are calculated, and the administrator may thus choose only relevant subtests.

The D-KEFS was normed on a stratified sample of 1,750 typically-developing children, adolescents, and adults in the United States (US) based on 2000 US census data (Delis et al., 2001). The sample ranged in age from 8 to 89 years old, and at least 75 people were in each of the 16 age groups used to create age-based norms. The stratification method considered age, sex, race/ethnicity, geographic region, and years of education. It provides normed data based on age, thereby accounting for developmental abilities. D-KEFS subtests were demonstrated to have reliabilities comparable to those of other available tests of EF (Delis, Kramer, Kaplan, & Holdnack, 2004). No comparisons to measures of unitary EFs were conducted. However, correlational analyses for each subtest relative to other subtests within the D-KEFS for typically-developing individuals were conducted, suggesting that the low correlations between subtests indicate that each reflects a distinct executive function, and that the relative impact of each executive function differs across age groups (Delis et al., 2001). For the TMT specifically, a moderate to high split-half reliability of 0.57-0.81 is reported across task components.

Validity of subtests within the D-KEFS was reported through intercorrelations across measures, correlations between the D-KEFS and other measures of EF, and through reports on studies with clinical populations (Delis et al., 2001). Although no studies of the TMT's relationship between other measures of CF were reported, the test manual reports that many subtests were developed through modification of pre-existing measures that had established validity (Delis et al., 2001).

The TMT was selected for the purpose of the present study. This subtest was administered as per the standardized instructions detailed in the administration manual. Time spent on each component of the subtest is the raw data used to obtain norm-referenced scaled scores (*M*=10, *SD*=3).

The TMT is a measure of CF consisting of five timed components: TMT1: Visual Scanning; TMT2: Number Sequencing; TMT3: Letter Sequencing; TMT4: Number-Letter Switching; and TMT5: Motor Speed. On TMT1, the individual is asked to visually scan a page containing circled numbers, and cross out each number three they detect. On TMT2, the individual is asked to look at a page of scattered circled numbers and connect all the numbers in sequence. TMT3 is similar to TMT2, but uses letters. The individual is asked to look at a page of circled letters and connect them in sequence. TMT4 is the measure of interest as it requires the individual to perform the same task as in TMT2 and TMT3, but to make their connections alternate from numbers to letters in ascending order. The primary measure of performance on this task is derived from a comparison of the individual's performance on TMT4 to their performance on TMT2 and TMT3. TMT5 is a task of motor speed where the individual is asked to simply connect a series of empty circles by way of tracing a dotted line as quickly as they can. TMT2 and TMT3 are used as baseline measures, and contrasted to performance on TMT4. The difference between the time taken to complete tasks TMT2 and TMT3, and TMT4 is used to reflect CF. A larger difference in time taken to complete TMT4 relative to TMT2 and TMT3 is indicative of impairment in CF (Delis et al., 2001).

Wisconsin Card Sorting Test. The Wisconsin Card Sorting Test (WCST; Heaton, 1993) is a widely used measure of EF that has been previously used in examinations of RB and CF in adults with ASDs (Lopez et al. 2005; South et al. 2007). It produces a measure of perseverative errors that reflects stuck-in-set perseveration. The task involves shifting from a prepotent, or previously reinforced, set of rules to a new set of rules that the participant must determine and shift to. This task provides normative data for ages 7 to 89 (Heaton, Chelune, Talley, Kay & Curtiss, 1993). Heaton et al., report that construct validity for the WCST as a measure of EF is supported by comparisons to other EF tasks, and that evidence from neurologically impaired populations suggests that the WCST is a valid measure of EF (1993). A study of children with ASDs found test-retest generalizability coefficients above .90 over an interval of two and a half years (Ozonoff, 1995). A computerized version of the test was used to increase consistency across administrations and reduce the influence of extraneous variables (Shu et al., 2001).

The 128-card WCST was computer administered with each participant in the following manner. Each participant was shown a laptop computer screen that displayed four key cards, each depicting a shape that varied in three features (colour, shape, and number). Each of these features had four variations, with numbers ranging from one to four, colours including red, green, yellow, and blue, and shapes including stars, circles, crosses and triangles. For example, one of the four key cards may display three yellow circles, another may display two blue stars, and so on in differing variations. At the bottom of the screen a simulated deck of cards appears, and the top card on the deck is revealed. The participant is asked to correctly match this top card to a key card, but is not told a strategy to use. Once the participant matches a card, the computer screen announces whether their placement is “right” or “wrong” and displays “right” or “wrong” on the screen according to a principle that is unknown to the subject, and that may change without warning. Thus, their task is to match cards correctly as much as possible, and to shift matching strategy when they are given negative feedback. This sequence is repeated over 128 trials. The matching principle chosen by the computer program is based on one of the three categories being chosen. If and when the participant has correctly matched ten consecutive cards, the matching principle is switched automatically (e.g., from number to shape) without notifying the participant. The matching principle changes among the three categories after the required number of correct responses until 128 trials are completed. The WCST was administered to each participant by a trained graduate student using a script (See Appendix A).

Performance on the WCST results in six principal scores, some of which are derived from other scores. Card sorts that match an incorrect category are classified as errors, and those that match the previously correct category are classified as perseverative errors. A standard score reflecting the amount of perseverative errors made was the primary dependent variable of interest in this study as it is a reflection of set-shifting and inhibitory ability. Thus, impairment in set-shifting on this task is evidenced by significant perseveration errors in responding, following a change in principle despite negative feedback (*M*=100, *SD*=15).

Parental questionnaires. In addition, one parent of each participant was asked to complete the YSIS, as well as a questionnaire on participant developmental history and demographics. Completion of these questionnaires within the larger study took the parent approximately one hour.

Procedure

Participants were recruited at a number of levels in Calgary, Alberta through a range of methods. Posters advertising the project were posted around the university campus, and distributed to interested elementary schools within the Calgary Board of Education. Brochures about the study were handed out to interested parents at presentations delivered by the project's principal investigator. The project was advertised on a morning segment through a local television station, and in local newspapers and magazines. As this study was contained within a larger research project, potential participants were informed about a strengths-focused study of children with high-functioning ASDs.

Potential participants who expressed interest in the study were contacted to arrange two appointments at the University of Calgary for the child and his or her parents to complete the study components. Parents received a copy of the informed consent form for their review by email prior to their scheduled appointments. Parents were asked to once again review, and sign the informed consent form upon their arrival at the initial appointment. Once informed parental consent was acquired, the participants and their parent(s) were requested to complete an initial screening process involving the measures for inclusion criteria described above. Subsequent to this initial screening, eligible participants were invited to complete the components of the larger research project, which took approximately four hours altogether. The participating child completed the TMT and WCST with an examiner. To ensure that children were performing to their best ability, each examiner was experienced with administering standardized assessments to children with various needs and took breaks between measures to increase each child's energy level.

Parents were asked to independently complete several questionnaires. Additionally, an interviewer trained to use the semi-structured ADI-R interview interviewed parents to gather information regarding their child's RBs. Upon completion of the two appointments, the participating child received a small toy and the parents received a $25 gift certificate to thank them for their participation. Refreshments and snacks were also available for participating children throughout each session. Participating families were informed that they would have the opportunity to learn about the global results of the larger research project after its completion. Collected data was hand-and computer-scored, and scoring was double-checked by another researcher.

Statistical Analyses

One sample *t*-tests were used to compare the performance of the experimental group to normed population performance on measures of CF. An alpha level of .05 (two-tailed) was used to indicate significance for all statistical analyses.

Correlational analyses were performed to explore the relationships between data collected from parents regarding their child's RBs and data collected based on the child's performance on tasks of cognitive flexibility. Specifically, correlations were run between degree of stuck-in-set perseveration and time spent on higher-order RBs, as well as simple perseveration of a previous response and frequency and markedness of repetitive motor movements.

CHAPTER 3. RESULTS

Following data collection, data were entered into a database and analyzed through a statistical software program. There were no missing items on any of the measures. An examination of box-plots revealed no extreme outliers, and thus no data points were deleted or adjusted.

Descriptive Statistics

Cognitive flexibility. Means and standard deviations for the D-KEFS and WCST are summarized in Table 2. Performance on the WCST was measured as mean perseverative errors and resulted in a standard score of 95.9 (SD=19.7), which is within the average range. On the TMT4 contrast score of the D-KEFS, participants had a mean score of 8.43 (SD=3.4) which is also within the average range.

Repetitive behaviours. Means and standard deviations for the YSIS and ADI-R RB domain are summarized in Table 3. On the YSIS, parents reported that child participants devoted between 25-75% of time left on their own to their circumscribed interest (mean=4.43, SD=1.75) over their preschool and elementary school years. On the ADI-R, parents reported that they observed only occasional motor stereotypies (mean=1.71, SD=1.76) both in their child's past and current functioning. It is important to note that 43% of participants were reported to not have exhibited any whole-body, or hand motor stereotypies in their lifetime.

Research Question One

Cognitive flexibility. *T*-tests failed to reveal a statistically significant difference between performance on the WCST in children with HFASDs and the normed population through comparisons of their performance: WCST $t(20)=-.940$, $p>.05$, $\alpha = .05$. However, a significant difference was found on the performance of children with ASDs and the normed population on the D-KEFS TMT4 Contrast Score: D-KEFS TMT4 $t(20)=-1.84$, $p=0.049$, $\alpha = .05$. Table 2 contains the values for these comparisons.

Research Questions Two and Three

Relationship between repetitive behaviours and cognitive flexibility. Table 4 shows the Pearson product-moment correlations (two-tailed) between the measures of CF and RB. These analyses indicated no significant relationship ($r = -0.05$) between higher-order RBs, as measured through parental ratings on the YSIS and stuck-in-set perseveration, as measured by the WCST. Similarly, no significant relationship ($r = -0.09$) was found between lower-order RBs, as measured through the ADI-R and simple perseveration, as measured through the D-KEFS TMT4 Contrast score.

CHAPTER 4. DISCUSSION

Based on previous research of individuals with AD, it was hypothesized that children with HFASDs would display impairment in CF, and show a relationship between degree of RBs and lack of CF. Overall, children in this sample showed significant impairment in simple perseveration CF in comparison to the typically-developing norming samples; however, they did not demonstrate an impairment in stuck-in-set perseveration as measured by the WCST. Furthermore, no statistically significant relationship was found between either higher-order RBs and stuck-in-set perseveration or lower-order RBs and simple perseveration.

Results on Research Questions

Research question one. It was found that this sample of children with HFASDs demonstrated impaired performance on the D-KEFS TMT4, a task of simple perseveration and CF. This result is consistent with previous findings (Lopez et al., 2005; Rumsey & Hamburger, 1988). However, the current sample demonstrated no impairment in stuck-in-set perseveration as measured by perseverative errors on the WCST.

There are several possible explanations for these two contrasting findings. First and foremost, there is inconsistency around the measurement of CF across studies. While some studies simply use one measure and one score to reflect the same construct (South et al., 2007) several studies have created various composite scores of CF whereby scores from different measures are grouped together to create new variables (Lopez et al., 2005; Salimpoor, 2006), for example grouping scores from the WCST and TMT4 (Lopez et al., 2005).

There has also been inconsistency in the specific scores that are used as dependent variables to represent CF, likely because standardized tests of EF often provide an array of scores rather than one unitary measure of each process to allow the examiner to select the score that is most pertinent to their interests. An example relevant to the present study is that some researchers using TMT4 neglect to take the examinee's performance on the foundational tasks (i.e., TM1 letter switching and TM2 number switching) into account when performing analyses and use the time the examinee required to complete TM4 as the dependent variable (Lopez et al., 2005), rather than the Number-Letter Switching contrast score that is produced by the scoring algorithm of the D-KEFS. These inconsistencies make comparison across studies challenging, as amalgamating different scores and measures impacts how the underlying construct is then represented. This practice may contribute to the variability in results observed in the present study.

Additionally, the lack of impairment found on the WCST in the present study is comparable to Ozonoff's findings (1995) wherein impairment on the WCST was not as pronounced when the task was administered through a computer version rather than manually (Ozonoff, 1995). Whereas typically-developing individuals have been shown to perform comparably on both the traditional and computer-administrations of the test (Artiola i Fortuny, & Heaton, 1996; Steinmetz, Brunner, Loarer, Houssemand, 2010), children with ASDs did not perform significantly more poorly than typically-developing controls on the computer-administration of the task (Ozonoff, 1995). They continued to perform significantly more poorly than controls in the traditional, manual-administration format (Ozonoff, 1995). While it can be assumed that CF is still necessary for both the computer and manual administrations, it could be that the improvement in performance was due to the decrease in social demands from the computer administration (Ozonoff, 1995). This finding begs the question of which cognitive ability is actually impaired when children with ASDs show a deficit in the manual administration of the WCST? Interestingly, despite this apparent attenuation in performance relative to the control group, the within-group performance of children with ASDs did not improve significantly between the manual and computer administration (Ozonoff, 1995). Moreover, a more recent study of children with ASDs in Taiwan found that CF impairment was evident using a computer-administration of the WCST relative to controls (Shu et al., 2001). Given these mixed findings, these studies must be replicated before any conclusions regarding the construct validity of the WCST are drawn based on these findings.

Research question two. No correlation was found between time spent on higher-order RBs and stuck-in-set perseveration. This is in contrast to previous findings with adults with ASDs (Lopez et al., 2005; South et al., 2007). A possible explanation for this lack of observed correlations is that the pattern of CF in this sample was atypical relative to previously researched samples (Lopez et al., 2005). Foremost, performance on the WCST was better than expected and was not significantly poorer than that of the typically-developing normed population. While no impairment on this task relative to typically-developing children has been found in one study of children with AS (Nyden et al., 1999), this is in contrast to a multitude of other studies that have found an impairment in children with ASDs (Ozonoff et al., 1991; Ozonoff & McEvoy, 1994; Prior & Hoffman, 1990; Shu et al., 2001; Szatmari et al., 1990). The present findings should thus be considered with caution.

Research question three. No correlation was found between degree and intensity of lower-order RBs and perseverative responding; however, the low correlation observed was likely largely accounted for by the very limited range of values observed in the RB domain of the ADI-R. As a large proportion (42%) of participants was reported not to have exhibited any motor mannerisms or stereotypies, this correlation is likely deflated (Tabachnik & Fidell, 2006). Indeed, a smaller proportion of the current high-functioning sample of children exhibited lower-order RBs than has been previously observed (South et al., 2005). The difference in lower-order RBs in the current sample may be due to a lack of sensitivity on the ADI-R or to the retrospective nature of the interview.

Furthermore, and similar to the measurement of CF, inconsistency exists in the measurement of RBs, as researchers employ different measures, different scores, or various composites of scores to capture the wide range of RBs exhibited in ASDs (Lopez et al., 2005; South et al., 2007). In the present study, both the YSIS and ADI-R were used to capture various aspects of RB, but did not capture other aspects of RB such as insistence on routines or sameness in the environment so as to maintain specificity to the lower- and higher-order types of behaviours. This inconsistency is undoubtedly influenced by a lack of sensitivity in available RB measures to the variety and extent of RBs displayed by children with ASDs as no single measure is currently available that thoroughly assesses both lower- and higher-order RBs in individuals with ASDs (South et al., 2007).

Limitations

Several limitations are apparent in the present study. The most prominent limitation is the limited sample size. Due to the specificity of the age and diagnosis of the examined population, as well as the intensive nature of the data collection, obtaining a larger sample size was difficult. This limitation affected both the robustness and the variety of statistical analyses that were performed.

The representativeness of the sample of the general population of children with ASDs may also have been limited by a number of factors. Specifically, participation in this study required a substantial commitment of time, travel, and energy for both the children who participated and their parents. Additionally, it was largely limited to participants who resided in the Calgary region, and was difficult to access for participants who lived in more rural areas. These factors likely influenced the characteristics of the participant sample and thus limits the generalizability of the findings of this study.

Additionally, this study did not control for comorbid conditions such as anxiety or ADHD that may have impacted the performance of participating children on measures of cognitive flexibility. ADHD could have negatively impacted a participant's ability to focus on tasks or inhibit impulsive responding (American Psychiatric Association, 2000), thus undermining their potential performance. Different types of anxiety disorders could also have negatively impacted the performance of participants. For example, social anxiety could impact a child's ability to engage in a task in front of the examiner (Wittchen, Stein & Kessler, 1999), and test anxiety could make a participant hypersensitive to feedback (Beidel & Turner, 1988), such as is incorporated into the WCST. Despite efforts to ensure optimal cooperation and effort, children with comorbid ADHD or anxiety may have performed worse than their peers with ASDs alone due to the impairments inherent to these conditions.

Lastly, no typically-developing control group of children was used in this study. While normed data for both WCST and D-KEFS measures of CF allowed for comparisons to the performance of typically-developing children, no such data was available for measures of RB. As both lower- and higher-order RBs occur at very low frequencies in the typically-developing population around the sampled age range, no comparison data from typically-developing children on these behaviours was collected.

Future Directions and Implications

Future research examining RB and CF in the population of individuals with HFASDs should expand on the current study by gathering data from a larger and more diverse sample of participants, which could result in more robust results and more readily inform the theoretical understanding of RB as stemming from executive dysfunction. Future studies may examine this relationship by measuring RBs in a more continuous manner rather than dividing this behaviour into higher- and lower-order components. Such an examination will require the development of a comprehensive instrument to measure the variety of RBs exhibited in ASDs. As some RBs are best measured through frequency and duration and others through the degree of impairment they cause (South et al., 2007), developing such a measure may be challenging. Additionally, future studies should take into account emerging evidence about developmental changes in RBs (Chowdhury, Benson, & Hillier, 2010) by measuring RBs within limited time and age spans.

Furthermore, future studies should include lower-functioning individuals with ASDs so as to evaluate the EF theory and Turner's (1997) hypothesis across the spectrum. It has been suggested that perseverative ability is related to verbal IQ, and this variable could be co-varied in samples with varied levels of cognitive functioning (Hill, 2004).

Additionally, future research should expand on the present study by employing different neuropsychological measures of CF. It has been suggested that scores derived from the WCST are not specific to the cognitive process of CF as successful performance also relies on other cognitive processes (Ozonoff, 1995). For example, selective attention is required to attend to the relevant aspects of the stimuli, and encoding and understanding of feedback is required to shift set (Ozonoff, 1995). It is therefore suggested that differential analysis of component skills is not facilitated by the WCST (Ozonoff, 1995). The more recently developed Intradimensional-Extradimensional Shift (ID/ED shift) task of the Cambridge Neuropsychological Test Automated Battery (Hughes et al., 1994), is a task of CF that is presented in multiple stages, thus allowing more specific analyses than through the WCST (Hill, 2004). Using this measure to further dissect component processes and cognitive impairments in this population may aid in linking biological and behavioural factors in ASDs.

This study examined the relationship between RB and CF in children with HFASDs. The overall outcomes of this study are somewhat inconclusive. No relationship was found between parental reports of higher-order RB and child performance on a task of stuck-in-set perseveration. Furthermore, no relationship between parental reports of lower-order RB and child performance on a task of simple perseveration was found. Given that the current study participants showed an atypical pattern of both lower-level RBs and stuck-in-set perseveration, both these findings should be considered with caution. Nonetheless, this study offers preliminary developmental data to add to previous studies of this relationship (Lopez et al., 2005; South et al., 2007). It is hoped that a larger sample size can elucidate the present findings.

APPENDIX A: WISCONSIN CARD SORTING TEST EXAMINER SCRIPT

"This test is a little unusual because I am not allowed to tell you very much about how to do it. You will be asked to match each of the cards that appear here" (point to the first response card at the bottom center of the screen) **"to one of these four Key cards"** (point to each of the stimulus cards at the top of the screen).

"Using the mouse, point to the Key card" (using the mouse point to each of the stimulus cards at the top of the screen) **"that you believe matches this card"** (point again with your finger to the first response card at the bottom center of the screen). **"Once you have made a choice, press either the left or right mouse button to make your selection."**
"The computer will place your card under the Key card that you select, and a new card will appear at the bottom of the screen. If you wish to change your answer before the card stops moving, immediately click on the Key card or anywhere on the screen a second time. You will then be permitted to select again. However, you may not change your answer after the card stops moving. If this happens, don't try to click on the Key card a second time; just go on to the next item.

"I cannot tell you how to match the cards but the computer screen will show you each time whether you are right (correct) or wrong (incorrect). I will also say the same word the computer shows on the screen, 'right (correct) or wrong (incorrect)."" Or "The computer will also say the same word it shows on the screen, 'right (correct)' or 'wrong (incorrect)'. If you are wrong simply try to match the next card correctly, and then continue matching the cards correctly until the test is over. There is no time limit on this test. Are you ready? Let's begin"

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Table 1: Demographic and Clinical Information

	Mean	SD
Age (years)	10.2	1.6
Male (number)	20	n/a
Diagnosis (number)	n=21	
AD	6 (29%%)	n/a
AS	11 (52%)	
PDD-NOS	3 (19%)	
VIQ	117.38	14.46
PIQ	119.86	14.87

Table 2: Descriptive Statistics for Cognitive Flexibility

	M	SD	p
WCST Perseverative Errors	95.9	19.7	>.05
D-KEFS Trail Making 4 Contrast Score	8.43	3.44	0.049

Table 3: Descriptive Statistics for Repetitive Behaviours

	M	SD
ADI-R Total Repetitive Behaviours Ever displayed	1.71	1.76
YSIS Amount of Time Spent on Circumscribed Interest Ever	4.43	1.75

Table 4: Pearson-product moment correlations for CF and RB

Variable	YSII Amount of Time Spent on Circumscribed Interest Ever	ADI-R Total Repetitive Behaviours Ever Displayed
WCST Perseverative Errors	-0.05	—
D-KEFS Trail Making Test Contrast Score	—	-0.09