Title

From Subtypes to Phenotypes: Discovering the Clinical Predictors of RRB Profiles in ASD

Permalink

https://escholarship.org/uc/item/4tf534qn

Author

Berry, Kathleen Charise

Publication Date

2017

Peer reviewed|Thesis/dissertation

UNIVERSITY OF CALIFORNIA

Los Angeles

From Subtypes to Phenotypes:

Discovering the Clinical Predictors of RRB Profiles in ASD

A dissertation submitted in partial satisfaction of the

requirements for the degree Doctor of Philosophy

in Education

by

Kathleen Berry

2017

ABSTRACT OF THE DISSERTATION

From Subtypes to Phenotypes:

Discovering the Clinical Predictors of RRB Profiles in ASD

by

Kathleen Berry

Doctor of Philosophy in Education

University of California, Los Angeles, 2017

Professor Connie L. Kasari, Chair

Autism spectrum disorders (ASD) are defined by impairments in social communication skills and the presence of restricted and repetitive behaviors (RRBs). Research has primarily focused on the measurement and influence of social communication deficits, despite the impact RRBs can have on daily functioning for individuals with ASD and their families. To date, age and cognitive functioning have been the focus of examination relating RRBs to individual characteristics. There is a gap in our understanding and consistency of measurement tools, conceptualization of RRB subtypes and understanding of the relationship between individual characteristics and RRBs in individuals with ASD.

In the current study, parent reported rates of RRBs in 2,856 individuals with ASD ranging in age from 4 to 18 were examined in order to both characterize the RRB types measured

and for co-occurrence of other factors, such as coping skills, anxiety and hyperactivity. Using a standardized parent report of RRBs, factor analytic results indicate that there are 5 distinct RRB types captured which include, hand/body mannerisms, rigidity, perseverative interests, compulsivity, and self-injurious behaviors. Next, a cluster analysis revealed 5 distinct phenotypic cluster profiles of RRB presentation, with each cluster encompassing various intensities of each of the 5 RRB subtypes. Lastly, a multinomial logistic regression analysis (MLR) was run with age, IQ, severity of ASD symptoms as well as standardized values of hyperactivity, anxiety and coping skills as predictors of phenotypic profile membership. Most notably, ASD symptom severity scores and nonverbal IQ did not significantly predict profile membership; rather hyperactivity, anxiety and coping skills were the significant predictors of RRB profile membership. In particular, anxiety was the strongest predictor of membership when comparing the optimal "All Low" RRB profile to each of the other phenotypic profiles.

Results from this study have uniquely exhibited the limitations of using an individuals' IQ, ASD symptom severity scores or age as the only characteristics in adequately predicting RRB presentation profile; contrary to most literature on the topic. This discrepancy across studies highlights the importance of using a standardized measure to define and quantify RRBs across studies. Further, results of the current study expand our understanding of the potential characteristics and developmental domains that have the most influence on RRB presentation. Although beyond the scope of the current study, these findings have significant implications in understanding potential underlying mechanisms related to RRB presentation and the functions they may serve for individuals with ASD, which may lead to improved treatment approaches to indirectly influence RRBs.

The dissertation of Kathleen Berry is approved.

Jennie Grammer

Shafali Jeste

Jeffrey Wood

Connie L. Kasari, Committee Chair

University of California, Los Angeles

2017

TABLE OF CONTENTS

LIST OF FIGURES

LIST OF TABLES

ACKNOWLEDGEMENTS

I first want to thank the countless families that I've had the pleasure of meeting and working with through ASD intervention research studies around the country. You have all made countless sacrifices to help researchers understand and improve the lives of your children, which will carry on to future generations of children with special needs. I am honored to have been welcomed into your homes and lives with such open arms and I am in awe of the unwavering strength and hope I've seen. Without you, this work and others like it would not be possible!

Next, a huge heart felt thank you to my lab mates and fellow grad school crew who have gone through this entire journey with me and have been an incredible source of support, encouragement and provided perfectly timed comedic relief. I have been lucky to have an amazing group of friends to work along side over the past 5 years. I owe this final product to the tireless dedication of my lab mate, class mate, friend, and designated life coach, Dr. Alex Sturm. Without you, this would not be possible!

Of course, a huge debt of gratitude to my friends and family- I appreciate your patience, support and encouragement through a process that has been demanding at times and your unwavering love has made it possible to forge ahead! I have to thank my big brother, JT- thank you for *making* me strong and resilient over the years ☺ and for bringing 3 sources of pure joy and love into my life! Lauren, there are no words for the love and gratitude I have for you and for ALL the amazing qualities that you've brought to our family- so proud to call you my sister. Dad, *thank you* for your unconditional support, love, patience and understanding during the months of unanswered calls, I owe you!

Most of all, I owe every achievement, every accomplishment and every mountain of adversity I am able to climb to my mom. She lived in a bold, selfless, unconditionally loving and brave way and with such grace…. *no matter what.* I will continually and tirelessly reach for the remarkably high bar she set for me in what dedication truly is. I can only hope to leave a legacy that stands the test of time as she did.

Kathleen Charise Berry

EDUCATION

2017   Ph.D. Candidate Human Development and Psychology in Education.
            University of California, Los Angeles

2008   B.A. Psychology (Minor in Child Development)
            California State University, Fullerton

ACADEMIC TEACHING EXPERIENCE

2016   Teaching Assistant. Winter 2016. *Autism: Mind, Brain, and Education.*
            University of California, Los Angeles

CONFERENCE PRESENTATIONS

Berry, K., Panganiban, J., Shih, W., & Kasari, C. (May 2016). *Characterizing Communication in Verbal and Minimally Verbal Preschool Children with ASD.* Poster presented at the International Meeting for Autism Research. Baltimore, MD.

Berry, K., Harrop, C., Huynh, L., & Kasari, C. (May 2015). *Repetitive and Restricted Behaviors in Infants at Risk for ASD: Comparing Caregiver Report and Observational Measurement.* **Oral presentation** at t he International Meeting for Autism Research. Salt Lake City, UT.

Gulsrud, A., Carr, T., Huynh, L., Berry, K., Paparella, T., & Kasari, C. (May 2015). Poster presented at the International Meeting for Autism Research. Salt Lake City, UT.

Berry, K., Gulsrud, A., & Kasari, C. (May 2014). *Generalization of Engagement in the Classroom Following a Parent-Mediated Intervention for Toddlers with ASD.* Poster presented at the International Meeting for Autism Research. Atlanta, GA.

Berry, K. & Kasari, C. (January 2012). *Maximizing Communication in Play: The Role of Parent Engagement.* Poster presented at the Special Education, Disabilities, and Developmental Risk (SPEDDR) conference. Santa Barbara, CA.

PUBLICATIONS

Kasari, C., Gulsrud, A., Paparella, T., Hellemann, G., & Berry, K. (2015). Randomized comparative efficacy study of parent-mediated interventions for toddlers with autism. *Journal of Consulting and Clinical Psychology, 83(3),* 554-563.

INVITED LECTURES

2015   "Teaching to Play or Playing to Teach". Pediatric Minds Institute. Torrance, CA.

AWARDS

2014   Graduate Summer Research Mentorship Fellowship
2013   University Fellowship
2013   Graduate Summer Research Mentorship Fellowship
2012   University Fellowship, Robert Levine Scholarship

EMPLOYMENT

2011-2016	Graduate Student Researcher. Kasari Research Laboratory. Center for Autism Research and Treatment. **University of California, Los Angeles**. Dr. Connie Kasari
2010-2011	Research Assistant and Classroom Instructor (Early Achievements classroom). **Kennedy Krieger Institute.** Baltimore, Maryland. Dr. Rebecca Landa
2010- 2011	Behavioral Clinician in Verbal Behavior Therapy. **Trellis Services, Inc**. Baltimore, Marlyand
2008- 2010	Research Assistant. Useful Speech Study/JASP Project. **University of North Carolina, Chapel Hill.** Dr. Linda Watson
2008	Behavioral Clinician. **Koegel Autism Center. University of California, Santa Barbara.** Drs. Robert and Lynn Koegel, Dr. Rosy Fredeen

CLINICAL SKILLS & TRAININGS

Early Interventions	Discrete Trial Training (DTT), Pivotal Response Training, Joint Attention Symbolic Play Engagement and Regulation (JASPER), Verbal Behavior, Sensory Integration Therapy, Enhance Milieu Teaching (EMT), introduction of Picture Exchange Communication System and Augmentative and Alternative Communication (AAC) devices
Assessments	Autism Diagnostic Observation Schedule (research reliable), Classroom Observation Rating Scale, Communication and Symbolic Behavior Scales (CSBS), Differential Ability Scales (DAS), Early Social Communication Scales (ESCS), Language Sample, Mullen Scales of Early Learning, Peabody Picture Vocabulary Test (PPVT), Preschool Language Scale- 4th Edition (PLS-4), Leiter-Revised, Receptive-Expressive Emergent Language (REEL), Structured Play Assessment (SPA)

Preface

Restricted and repetitive behaviors (RRBs) are a core diagnostic feature of autism spectrum disorders (ASD) (*Diagnostic and Statistical Manual of Mental Disorders,* 5th ed. (DSM-5); American Psychiatric Association, 2013). However, in relation to their diagnostic counterpart of social communication deficits, RRBs have received much less attention in ASD research (Leekam, et al., 2011; Bishop, et al., 2013; Bodfish, Symons, Parker, & Lewis, 2000; Elison, et al., 2014). RRBs include an extensive range of behaviors that include stereotyped movements, compulsions, repetitive use of language, repetitive manipulations of objects, severe attachment to objects, insistence on sameness, repetitive self injurious behaviors, and narrow circumscribed interests (Bodfish, 2007; Bodfish, Symons, Parker, & Lewis, 2000). This wide array of behaviors are all linked by the quality and frequency of repetition, rigidity, and inflexibility at which these behaviors occur throughout the life span for individuals with ASD.

The aim of this study was to explore the phenotypic patterns in which RRBs are manifested in individuals with ASD. After characterizing the patterns of RRBs, the influence that hyperactivity, anxiety and coping skills have on predicting RRB profile membership was explored.

INTRODUCTION

Although RRBs are a defining feature of ASD, they are not exclusive to the disorder and are present in typically developing children and in children with developmental delays (DD) (Thelen, 1979, 1981; Evans, et al., 1997; Watt, et al., 2008; Damiano, et al., 2013). Repetitive behaviors exhibited in early development are thought to be important mechanisms for learning processes in the acquisition and mastery of skills such as motor control and actions with objects (Thelen, 1979, 1981). Typically developing infants and toddlers exhibit a variety of RRBs such as repetitive motor actions, rigidity in routines, repetitive manipulation of objects and significant attachment to certain items (Thelen, 1981). Understanding what forms and functions RRBs serve in the typically developing population will better inform our understanding of how these behaviors may differ for individuals with ASD.

RRBs are now understood to be a continuum of behaviors that serve various functions throughout development and may occur frequently within typical infant and toddler development and vary based on children's skill acquisition and mastery level (Leekam, et al, 2007; Thelen, 1979, 1981; Evans, et al., 1997). Since repetition of actions serve the purpose of skill acquisition and mastery, as development progresses and mastery of skills is attained, RRBs reduce overtime in typically developing children (Thelen, 1981; Evans, et al., 1997; Wolff, et al., 2014). Specifically, studies have found consistent patterns of certain repetitive behaviors present in the first year, increase until roughly the age of three and start to decline around the fourth year (McDonald, et al., 2007). This reduction in repetitive behaviors over time has been found in studies using parent report measures of children in the first four years of life (Evans, et al., 1997), as well as observational coding of repetitive behaviors in young children (Harrop, et al., 2014).

Studies involving early developmental behaviors face the obstacle of the fluctuations that naturally occur in development, making the measurement of behaviors an important factor to consider. Specifically for RRBs, the type(s) of repetitive behaviors captured, environmental influences such as location or caregiver presence, and the measurement tool used can all influence results. Thelen (1981) examined body stereotypies of 20 infants in the first year using live behavioral observations across contexts such as feeding, interactions with caregivers and while the infants were interacting with objects. Thelen collected data on bouts of rhythmical body movements including movements of the legs (ex.; single-leg kick, alternate-leg kick, foot rub, etc.), torso (ex.; sit rock, stand bounce, sit sway, etc.) and arms (ex.; wave, bang object, arm sway, arm rotate, etc.). While stereotypies decreased over time, contextual triggers impacted the type of body stereotypies observed. Patterns in stereotypies were found to increase when the infants were interacting with their caregivers, and while in a heightened state of arousal. Understanding contextual dependency and fluctuations within the TD population will help to inform similar behaviors observed in children with ASD.

It has been posited that in the case of caregiver interactions, increased stereotypies may serve a communicative function; that is, kicking and bouncing rhythmically may serve communicative value to express pleasure and excitement. These differences found in frequency of stereotypies when a caregiver is present highlights the importance of measurement, and for many behaviors, parent report may be the most inclusive tool to better understand the presence of atypical behaviors across contexts.

Understanding the developmental pattern, frequencies and types of RRBs in typically developing children is informative in determining what constitutes atypicality of RRBs for children with ASD. The repetition of behaviors observed in typically developing infants and

toddlers exhibit overlap with the behaviors indicating developmental concern or impacting diagnostic outcomes in children with ASD. Unfortunately, information is somewhat limited on the presence, severity and developmental pattern of specific RRB subtypes longitudinally. More often, studies have examined the role of RRB presentation in differentiating children with ASD from other clinical or control groups. Findings have indicated that in contrast to the pattern found for TD children of RRBs peaking around 2 and dissipating by 4, children with ASD exhibit a continual increase in the frequency and severity of RRBs through late childhood (Moore & Goodson, 2003; Bishop, Richler, & Lord, 2006).

The organization, division and measurement of RRBs inevitably influence findings; such methodological inconsistencies have pervaded RRB research and limited understanding of complicated relationships between RRBs and other individual characteristics (Bishop, et al., 2006; Cox, et al., 1999; & Troyb, 2014). A variety of approaches have been taken to organize and operationally define RRBs in ASD. In order to build upon advances made in RRB research, it is essential to consider the methodological approaches taken (Mirenda, et al., 2010; Troyb, 2014).

Organization of RRBs

The decision to include or exclude certain RRB types significantly influence results, leading to inconsistencies across studies purely based on methodological limitations. Therefore, in order to sufficiently understand and improve this area of research, researchers must evaluate the organization, inclusion and exclusion of RRBs, which vary by study. As Troyb (2014) points out, findings vary based on the behavior(s) included, with inconsistent results between RRBs and, for example, functioning level, based on the RRB types examined. The most commonly used organization, definition, and measurement of RRBs will be described further.

4

Dichotomization of behaviors into low-level and high-level RRBs is one of the most common organizational approaches (Turner, 1999). An alternative label for these two categories are Repetitive Sensory Motor (RSM) behaviors for low-level RRBs and Insistence on Sameness (IS) for high-level RRBs (Cuccaro, et al., 2003; Bishop, et al., 2013; Richler, et al., 2010; Bishop, et al., 2006). This approach of dichotomizing RRBs is nearly identical, therefore low-level and RSM can be used interchangeably, and the same applies to high-level and IS behaviors.

Low-level RRBs include repetitive motor stereotypies such as hand flicking, body rocking, etc., stereotyped or repetitive speech vocalizations, and repetitive actions with objects such as spinning wheels, repetitively opening and closing containers, etc. These behaviors may also possess sensory components, implying a physiological function may be simultaneously served while children engage in certain RRBs (Lovaas, Newsom, & Hickman, 1987; Boyd, et al., 2009; Baranek & Bodfish, 2009). RRBs conceptualized as low-level are often associated with younger and lower functioning children, yet are also present in early typical development and in other developmental and psychiatric conditions such as Fragile-X syndrome, Rett syndrome and Tourette's syndrome (Thelen, 1979; Frith & Done, 1990; Turner, 1996; Evans, et al., 1997; Damiano, et al., 2013; Watt, et al., 2008).

High-level behaviors encompass behaviors such as intense preoccupation with a restricted interest, ritualized behavior patterns, and excessive adherence to routines with significant resistance to change (Turner, 1999). The most commonly described high-level RRBs include behaviors such as rituals and routines, which make up insistence on sameness (IS) behaviors (Szatmari, et al., 2006). These rigid behavioral patterns were documented in the original description of ASD as a staple of the unique features of the disorder (Kanner, 1943). High-level behaviors are commonly associated with older and higher functioning children, when

children are more likely to be able to communicate and share intense and perseverative interests, commonly referred to as circumscribed interests, with others (Esbensen, et al., 2009; Richler, et al., 2010). It is possible for multiple subtypes, that is, across high and low level RRB classifications to take place simultaneously, further complicating the measurement and recognition of each RRB presentation (Leekam, 2011).

Despite the popularity of this simple shorthand to group behaviors, it has been cautioned that this approach is too broad and may obscure important differences between the many types of RRBs (Turner, 1997; 1999; Leekam, et al., 2011). RRBs have been organized in a number of ways; however differences among how studies categorize behaviors reduce comparability of results. Results are dependent upon the measurement tools used to organize and define various RRB subtypes; therefore it is important to understand the strengths, weaknesses, inclusion and utility of common measurement tools in exploring patterns of RRBs among individuals.

The diagnostic criteria for autism spectrum disorder (ASD) has recently been altered to a dyad of impairments in the domain of social communication ability and the presence of atypical restricted and repetitive behaviors (RRB) (*Diagnostic and Statistical Manual of Mental Disorders,* 5th ed. (DSM-5); American Psychiatric Association, 2013). As the manual for defining and diagnosing a range of disorders, the DSM's definition of ASD symptomology is important to consider in conceptualizing definitions and measures of RRBs in ASD. The DSM-5 categorizes RRBs into four domains, children must manifest at least two of the following: (1) stereotyped or repetitive speech, motor movements, or use of objects; (2) excessive adherence to routines, ritualized patterns of verbal or nonverbal behavior, or excessive resistance to change; (3) highly restricted, fixated interests that are abnormal in intensity or focus; and (4) hyper- or hypo- reactivity to sensory input or unusual interest in sensory aspects of their environment.

Figure 1. Common Conceptualizations of RRB Subtypes
(From *Joseph, et al., 2013*)

Turner (1999)	Lower Order RRBs			Higher Order RRBs		
RSM and IS terms (Richler et al., 2010)	Repetitive Sensory Motor (RSM)			Insistence on Sameness (IS)		
Lam & Aman (2007) RBS-R Algorithm	Self-Injurious Behavior	Stereotyped Behavior	Restricted Behavior	Compulsive Behavior	Ritualistic/Sameness Behavior	
Bodfish et al. (2000) RBS-R Algorithm	Self-Injurious Behavior	Stereotyped Behavior	Restricted Behavior	Compulsive Behavior	Ritualistic Behavior	Sameness Behavior
Mirenda et al. (2010) RBS-R Algorithm	Self-Injurious Behavior	Stereotyped Behavior and Restricted Behavior		Compulsive Behavior, Ritualistic Behavior and Sameness Behavior		

Measurement of RRBs

The methodological inconsistency across studies is often due to the current lack of a consistent and universal measurement of RRBs. Measurement tools dictate the operational definitions employed, methodological variability in data and analysis between studies, and results found between RRBs and related characteristics (Rutter, 1996; Lewis & Bodfish, 1998; Turner, 1999; Leekam, et al., 2011). Parent reports have been the most prevalent form of measuring RRBs in children with ASD, which vary across studies.

Autism Diagnostic Interview- Revised (ADI-R; Rutter, et al., 2003). The ADI-R is a standardized semi-structured interview utilized to diagnose ASD and measure symptom severity. The ADI-R is a parent report measure designed to capture developmental history as well as current symptom presentation of individuals with ASD. The three domains the ADI-R addresses are: (1) language and communication, (2) reciprocal social interaction, and (3) restricted,

repetitive, and stereotyped behaviors. Each of the three domains has a cut-off score, providing a

diagnostic algorithm, which is accurate in differentiating an ASD diagnosis from other disorders.

Even though the ADI-R is not intended to independently and all-inclusively measure RRBs, a

number of studies have characterized RRBs in individuals with ASD using the RRB subscale of

the ADI-R (Cox, et al., 1999; Moore & Goodson, 2003; Bishop, Richler & Lord, 2006; Honey, et

al., 2007; Richler, Bishop, Kleinki & Lord, 2007).

The RRB domain of the interview includes information on both what the child has

displayed in the past as well as what behaviors the child is currently exhibiting. Most commonly,

studies examining RRBs using this measure exclusively use the "current" items only (Bishop, et

al., 2006; Richler, Bishop, Kleinki & Lord, 2007; Cox, et al., 1999). Notably, evidence for the

dichotomy of high and low-level RRBs were derived from studies that used the ADI-R as their

measurement of RRBs; therefore, it hasn't been unanimously established if that organizational

approach is applicable to all individuals with ASD, or if the dichotomy is due to the factor

structure of the ADI-R (Bishop, et al., 2013). Despite it's popularity, there is also methodological

concern when studies use a measure for multiple purposes; as the intended use for the ADI-R is

to measure a continuum of ASD characteristics and determine diagnostic eligibility. Therefore,

utilization of a measurement tool solely for the purpose of measuring RRBs with established

validity, reliability and inclusion of all subtypes of RRBs would likely produce a stronger tool

for quantifying RRBs.

Repetitive Behavior Scale- Revised (RBS-R; Bodfish, et al., 2000). The RBS-R is a

questionnaire that was designed for the purpose of exclusively measuring a variety of RRBs. The

measure includes 43 items that are rated on a four-point Likert scale (ranging from 0 to 3) across

6 subscales, which were conceptually derived and reported by the primary caregiver. The

original subscales include: stereotyped behavior; self-injurious behavior, compulsive behavior, ritualistic behavior, sameness behavior, and restricted behavior. Since it's conception, several factor analytic studies have been conducted with the RBS-R with varying results, implying that the original factor structure of 6 subscales is not statistically supported based on these results (Lam & Aman, 2007; Esbensen, et al., 2009; Mirenda, et al., 2010; Bishop, et al., 2013).

Lam & Aman (2007) were the first to independently explore the factor structure of the RBS-R. They examined data from 307 participants and explored a large age range of individuals with ASD (3- 48 years old). Results indicated that the RBS-R provides five factors, which overlap with five of the six original scales; the Ritualistic subscale, originally proposed by Bodfish, et al. (2000), was the only scale not included in the new factor solution. This finding was supported in a subsequent study of 712 individuals ranging from 2 to 62 years old, which explored the five-factor model (Esbensen, et al., 2009). Additionally, Mirenda, et al. (2010) used a confirmatory factor analysis to compare several different proposed structure models and found the best models were the Lam & Aman (2007) five-factor model and a three factor model (*see Figure 1*). Most recently, Bishop, et al. (2013) explored the relationship between ADI-R scores and RBS-R relating to the construct validity of using the RSM and IS dichotomy for RRBs in over 1,800 individuals with ASD. Results from the initial exploratory factor analysis (EFA) of the RBS-R were similar, with slight divergence in items factor loadings from previous investigations (Lam & Aman, 2007) and from the original RBS-R factors (Bodfish, et al., 2000). In consideration of the varying results across studies, further exploration of the RBS-R factors is warranted.

Organization and measurement of RRBs is a complicated undertaking, with inevitable influence on outcomes when examining the relationship between RRBs and other developmental

characteristics (Leekam, et al., 2011). Therefore, advances in understanding the complicated relationships between RRB presentation, chronological age, cognitive functioning, and other developmental skills have progressed more gradually. However, incremental advancement is logical given the complexity and difficulty in RRB measurement. Despite the complexity of RRB presentation and the numerous issues in organization and measurement described, careful evaluation of the phenotypic patterns and related characteristics found warrants further consideration.

Age and RRBs

The relationship between age and RRB presentation in ASD has most commonly been examined through the use of cross sectional data analysis (Esbensen, et al., 2009; Militerni, et al., 2002; Bishop, Richler, & Lord, 2006; MacDonald, et al., 2007). Researchers have found that that younger children with ASD exhibit higher frequency of *low-level* RRBs such as motor stereotypies and sensory related behaviors; whereas older, higher functioning individuals on the spectrum tend to exhibit more *high-level* RRBs, with reduction in low-level RRBs (Militerini, et al., 2002; Bishop, Richler, & Lord, 2006; Esbensen, Seltzer, Lam, & Bodfish, 2009). Specifically, Militerni, et al. (2002) found that toddlers (2-4 years old) exhibited significantly fewer RRBs than older children (7-11 years old); though, this was only true for sensory and motor RRBs (low-level RRBs), which were significantly less prevalent in the older group. However, the older children were not devoid of RRBs, they instead displayed more complex (high-level) RRBs such as routinized schedules and insistence on sameness.

The developmental trajectories of children with ASD are complex in their symptom presentation across time, further complicated by the manifestation of various types of RRBs. There have been several studies to examine RRB presentation overtime, with varying results

10

across studies (Moore & Goodson, 2003; Richler, et al., 2010; Harrop, et al., 2014). The most common finding in regards to age and RRBs has been an overall reduction overtime in RRBs, with a more significant decrease overtime in low-level RRBs such as repetitive object use or motor actions (Honey, et al., 2008; Moore & Goodson, 2003). Moore & Goodson (2003) followed children with ASD from two to four years old and found that overall rates of RRBs reduced, with low level RRBs significantly reducing, yet different RRB subtypes persisted in a more complex form. Whereas Honey, et al. (2008) examined preschoolers and found within a year there was a significant decrease in the severity of RRBs observed.

However, a more recent study, which utilized observational coding of RRBs in a preschool aged sample, found there was no significant change in any of the RRB subtypes coded across 13 months and three assessment time points (Harrop, et al., 2014). Further, Richler, et al. (2010) used the ADI-R to track change in RRBs over a period of 9 years and found that low-level sensory motor RRBs actually remained high. Taken together, these findings suggest that the developmental progression and transformation of RRBs overtime within individuals remain unclear, with the biggest influence being the sample population and measurement tools used (Leekam, et al., 2011; Harrop, et al., 2014; Richler, et al., 2010).

Cognitive Functioning and RRBs

Perhaps the most consistent finding in RRB research is the relationship between RRBs and intellectual functioning (Militerni, et al., 2002; Bishop, Richler, & Lord, 2006; Esbensen, et al., 2009; Kim & Lord, 2010; Ray-Subramaian & Weismer, 2012; Rao & Landa, 2014). The general consensus within the field is that children with more severe adaptive and cognitive impairments exhibit higher frequency, more intense, and more persistent RRBs (Gabriels, et al., 2005; Esbensen, et al., 2009; Bishop, et al., 2013). More specifically, children with lower

cognitive capacity exhibit the most frequent and severe low-level RSM RRBs, whereas children with higher cognitive capacity exhibit significantly less of the RSM behaviors (Militerni, et al., 2002; Bishop, Richler, & Lord, 2006; Esbensen, et al., 2009; Rao & Landa, 2014). However, these findings are nuanced, as there is not a singular and linear relationship between IQ and RRBs, and consideration must be given to the types of RRBs being measured. For example, in a study examining 830 children with ASD between 15 months and 12 years old with an average age of 5 years old, found that for many RRBs, a significant interaction effect was found between nonverbal IQ (NVIQ) and age (Bishop, Richler, & Lord, 2006). Specifically, in older children, NVIQ was strongly related to low-level RRBs such as hand and finger mannerisms. However, high-level RRBs like circumscribed interests were positively related to NVIQ. Bishop, Richler, & Lord (2006) used the ADI-R to examine the total as well as the individual types of RRBs, which included 13 types of behaviors considered to fall under the RRB umbrella. Another interesting finding from this study was the relationship between NVIQ and RRBs actually became stronger with increasing age, where children under the age of 3 showed no relationship between RRBs and NVIQ. Similarly, Kim and Lord (2010) found no association between NVIQ and RRBs in toddlers under two years old. Taken together, these findings further evidence the importance of measuring subtypes individually, as there are clear differences across RRB types in the relationship between cognitive ability and RRB presentation.

Findings across studies highlight the importance of examining specific subtypes of RRBs and the traits associated with them, as these traits may significantly impact the persistence or the possible reduction of RRBs overtime. For example, Ray-Subramaian and Weismer (2012) found that not only were receptive and expressive language skills significantly lower among children with higher rates of RRBs, but they also concluded that higher scores in both language domains

in 2-3 year olds could significantly predict a reduced rate of RRBs. Again, consumers of this area of ASD research must take into account the number of participants and age span included across studies. Most recently, a study examined children at three time points to determine if RRB presentation at 1-2 years old, and/or 3-5 years old can predict cognitive functioning, adaptive skills and ASD symptomology at 8-10 years old (Troyb, 2014). Results showed that increased severity of low-level RRBs (specifically sensory interests and repetitive motor movements) were significant predictors of lower cognitive and adaptive skills as well as a greater ASD symptom severity at age 8-10 years. This relationship was not found when examining whether RRBs in the first two years of life could significantly predict the same school-aged outcomes (Troyb, 2014). Despite the nuanced findings among studies regarding the specific relationship and influence RRBs have on cognitive performance, it is clear that future studies must consider and control for cognitive functioning when examining the relationship between RRBs and other clinical characteristics (Gabriels, et al., 2005).

Adaptive Functioning and RRBs

Children with ASD exhibit significant impairment in adaptive functioning skills that extend beyond their cognitive deficits (Liss, et al., 2001). It is important to note that adaptive functioning skills measure the ability of an individual to successfully function within their given environment, and studies have demonstrated greater deficits in adaptive functioning for children with ASD compared to age and IQ matched peers (Carpentieri & Morgan, 1996; Volkmar, et al., 1987). Several studies have examined the relationship between adaptive skills and RRB presentation in individuals with ASD (Szatmari, et al., 2006; Honey, et al., 2007; Mooney, et al., 2009; Mirenda, et al., 2010). Similar to the relationship between RRBs and IQ, results have varied based on the measures used and age of participants; however, it can be deduced that in

general, higher rates of RRBs are associated with lower adaptive functioning skills (****).
However, this finding has varied across studies based on the age and IQ of the participant (Liss,
et al., 2001; Gabriels, et al., 2005; Militerni, et al., 2002).

The relationship between adaptive skills and RRB presentation in ASD is complex and
has varied results based on the age and IQ of participants as well as the measures employed. For
example, Liss, et al. (2001) found that the relationship between adaptive functioning and RRBs
was dependent on the severity of adaptive skill impairment. Specifically, there was no significant
relationship between RRBs and adaptive functioning in lower functioning children with ASD;
yet the high functioning group exhibited a significant correlation between adaptive behaviors and
RRBs.

Few studies have examined adaptive functioning and RRBs using measures other than
parent report. However, in young children, self-regulation has been observed in interactions
between parent and child. As shown by Wetherby and Prizant (2002), children with ASD
exhibited lower proportions of well-regulated behavior bouts and higher incidences of RRBs
during parent child interactions. Theoretically, some have suggested that specific RRBs may be a
result of an emotional trigger for children with ASD. However, Militerni, et al. (2002) found that
most (71%) of the low-level RRBs observed in 2-7 year olds (n=121) were not reactive to a
particular emotional trigger. The remaining 29% of RRBs deemed to be reactive in nature
consisted of high intensity sensory behaviors, including self-injurious behaviors, motor RRBs
and sensory stimulation, which were all more common the younger participants (Militerni, et al.,
2002). This notion of an emotional trigger also highlights a theory that RRBs serve as a coping
strategy to regulate their state of arousal; however, there are currently not enough results or data

to full endorse this theory and further examination is needed (Barber, 2008; Leekam, et al., 2011).

ASD-Related Psychopathologic Traits

There are a number of common developmental and neuropsychiatric disorders that overlap in symptom presentation, and in some cases are determined to co-occur in children with ASD. Some of the most common are attention deficit hyperactivity disorder (ADHD) and anxiety disorders (Rao & Landa, 2014; Wilens, et al., 2002; Zandt, Prior & Kyrios, 2007; Ruzzano, 2012; Joosten, Bundy & Einfeld, 2008). There is limited knowledge about how these ASD-related disorders vary across the population and what impact the co-occurring conditions impact RRB manifestation as well as the impact on overall development, adaptive skills and other child characteristics.

Attention deficit hyperactivity disorder (ADHD). Attention deficit hyperactivity disorder (ADHD) is a neurodevelopmental disorder characterized by symptoms of inattention, impulsivity, and/or hyperactivity exhibited to a degree substantially beyond what is expected for developmental level (Wilens, et al., 2002). ADHD and ASD share overlapping symptoms such as issues with communication problems, issues with attention and the presence of restricted behaviors (Hattori, et al., 2006). Although the last version of the *Diagnostic and Statistical Manual of the American Psychiatric Association* (4th ed., *DSM-IV-TR*; American Psychiatric Association, 2000) prohibited a dual diagnosis of ASD and ADHD, preliminary evidence suggests that when these two disorders co-occur, the risk for increased severity of psychosocial issues intensifies (Yerys, et al., 2009). Such findings are in conjunction with a growing number of researchers reporting children who meet criteria for both disorders are evidence to suggest they can co-occur (Gadow, et al., 2006; Holtmann, et al., 2007; Yerys, et al., 2009).

The need for research to examine the dual presence of clinically significant ADHD symptoms in individuals with ASD has begun to be addressed (Rao & Landa, 2014). This study compared school-aged children 4 to 8 years old that included younger siblings of children with ASD (with ASD n=27 and without an ASD diagnosis n=75), children with ASD (n=35), low-risk controls (n=12) and children with language delay (n=13) to include reference points for functioning and skill level across groups. Results indicated that children with comorbid ASD and ADHD diagnoses had lower cognitive functioning, more severe social impairments, and greater delays in adaptive functioning than children with ASD only (Rao & Landa, 2014). There is a great need for continued exploration of the impact of co-occurring ASD and ADHD symptomology on children with ASD; specifically, how elevated levels of hyperactivity influence the presence and severity of RRBs in children with ASD.

Anxiety. The role of anxiety for individuals with ASD has been proposed to play a key role in the severity of RRBs, as the function of engaging in specific RRBs has been hypothesized to serve as a coping mechanism to reduce feelings of anxiety (Troyb, 2014; Carruthers, 1996). However, it should be noted that there is insufficient evidence currently to support this theory (Leekam, at al., 2011). Scientific evidence illustrating links between anxiety, ASD and RRBs is limited. However, there have been studies that indicate high levels of anxiety in the ASD population and even links to symptom severity increasing (Tonge, Brereton, Gray & Einfeld, 1999). The link between anxiety and ASD symptom severity is logical considering the need for routine, sameness and consistency to a severe degree. Interruption of those may result in increased levels of anxiety and intense stress often accompanied by outbursts when self-control is impaired. The theories accounting for the popular notion that anxiety and arousal states

significantly contribute to increased RRB severity for individuals with ASD still needs to be

explored (Leekam, et al., 2011).

Current Study

The present study aimed to explore the phenotypic presentation of RRBs and associated

characteristics for individuals with ASD between 4 and 18 years old. Previous studies have been

limited by measurement tools, limited age(s) included, as well as limited statistical power due to

smaller sample sizes; therefore, this study aimed to examine the forms of RRBs across age and

IQ, and to examine the impact of hyperactivity, anxiety, coping skills, and ASD severity on RRB

presentation in a large, well-characterized sample of individuals with ASD.

The first aim was to define the specific RRB subtypes derived from a factor analysis of

the Repetitive Behavior Scale- Revised (Bodfish, et al., 2000). Secondly, the RRB subtypes

derived from the factor analysis were used to cluster participants based on type and severity of

co-occurring RRB subtypes into phenotypic profiles.

The final aim of this study was to explore the role of clinical (ASD symptom severity,

anxiety and hyperactivity), cognitive (nonverbal IQ) and adaptive skills (coping) in predicting

phenotypic profile group membership. Researchers have yet to uncover the specific function/s of

RRBs; therefore, examination of the predictive power of individual clinical characteristics on

RRB phenotypes contributes to this area of research.

METHOD

Data Acquisition

The Simons Foundation Autism Research Initiative (SFARI) provides researchers with access to multiple databases of systematically collected clinical measures from families across the United States. In March of 2016, the Institutional Review Board (IRB) at the University of California, Los Angeles (UCLA) determined that a full review was not necessary for the principal investigator to submit the application to access several SFARI databases (IRB# 16-000314). In May 2016, SFARI granted access to two relevant databases; Simons Simplex Collection (SSC), and Simons Ancillary Collection. SFARI databases were designed for large-scale data sharing; therefore, the majority of cognitive and behavioral assessments analyzed in the current study were administered across each project.

The largest dataset, the Simons Simplex Collection (SSC) includes data from 2,644 probands collected across twelve university-based sites: Baylor College of Medicine, Children's Hospital Boston/Harvard Medical School, Columbia University, Emory University, McGill University, University of California, Los Angeles, University of Illinois at Chicago, University of Michigan, University of Missouri, University of Washington, Vanderbilt University and Yale University. Phenotypic data was collected from the primary participants (probands) aged 4 to 18 from simplex families, in which only one child (probands) had an ASD diagnosis, with no other family history of ASD. The Simons Ancillary Collection includes 256 families that failed to meet criteria for the SSC, most often due to a family member being diagnosed with ASD or DNA sample/s were not collected for a primary family member, thereby excluding them from joining the SSC database.

Participants

A total of 2,856 individuals with ASD between 4 and 18 years old were included in the current study. Participants were primarily male (83.4%) and Caucasian (75.9%). Inclusion criteria for the probands included meeting ASD diagnostic cutoffs on the Autism Diagnostic Observation Schedule (*ADOS*; Lord, et al., 1999) and the Autism Diagnostic Interview-Revised (*ADI-R*: Rutter, et al., 2003). Participants also had to cognitively perform at a non-verbal mental age of at least 24 months for 4-7 year olds and 30 months for participants 7 years and older. Families were excluded if the proband had significant hearing, vision, or motor problems. Also, any known genetic syndromes (e.g., Down Syndrome, Fragile X, Tuberous Sclerosis) or significant prenatal or birth complications precluded their participation.

Participating families were initially screened to confirm eligibility based on ASD diagnostic measures and nonverbal cognitive assessments administered by reliable assessors (either a psychologist or physician with appropriate credentials). The diagnostic measures and cognitive assessments were done within the same 6-month period. Data collection was in accordance with the requirements of the Institutional Review Boards at each of the university sites. Each proband completed between 4 and 6 hours total of direct assessments, with additional collection of parent interviews, questionnaires and family history.

Measures

Cognitive Performance Measures

Probands were administered one of four cognitive assessments based on their age and cognitive functioning level to derive their nonverbal IQ (NVIQ) score as well as verbal IQ (VIQ) score. The standardized cognitive measures included the Differential Ability Scale, 2nd Edition

(DAS-II; Elliott, 2007), the Mullen Scales of Early Learning (MSEL; Mullen, 1995), the

Wechsler intelligences scale for children, 4th Edition (WISC-IV; Wechsler, 2003), or the

Wechsler abbreviated scale of intelligence (WASI; Wechsler, 1999). The majority of probands

completed the DAS-II (89%), while 6% completed the MSEL, and the remaining 5% completed

either the WISC-IV or the WASI.

The Differential Ability Scales, 2nd Edition (DAS-II; Elliott, 2007). The DAS is a

measure of cognitive abilities and assesses both verbal and nonverbal skills separately. Scores

from the DAS are standardized, norm-referenced and provide separate standard scores for verbal

IQ (VIQ) and nonverbal IQ (NVIQ). Recently, a study examined the convergent validity of the

DAS and the Mullen Scales of Early Learning (MSEL) in 53 children with ASD and 19 children

with non-spectrum diagnoses and found that there is good convergent validity between the DAS

and MSEL with NVIQ and VIQ scores (Bishop, Guthrie, Coffing, & Lord, 2011). Further, the

DAS has been shown to have good convergent validity with the Wechsler Intelligence Scale for

Children, 3rd Edition (WISC-III; Dumont, Cruse, Price, & Whelley, 1996). The standard scores

for NVIQ were used in all relevant analyses.

The Mullen Scales of Early Learning (MSEL; Mullen, 1995). The MSEL is a

developmental assessment for young children as a measure of cognitive skills across five

domains: *visual reception, fine motor, gross motor, receptive language and expressive language.*

This cognitive assessment is intended for children from birth through 68 months; participants

who were unable to complete the DAS were administered the MSEL.

The Wechsler Abbreviated Scales of Intelligence- Second Edition (WASI; Wechsler,

1999) and *The Wechsler Intelligence Scale for Children- Fourth Edition* (WISC-IV*;* Wechsler,

2003). Roughly 5% of participants completed either the WASI or the WISC. David Wechsler

developed the WISC and WASI, as well as several other intelligence tests. The WISC is individually administered to children aged 6 to 16, and the WASI can be administered to individuals through age 90 (Wechsler, 1999; 2003).

Parent-Reported Measures

Aberrant Behavior Checklist (ABC; Aman & Singh, 1986). The ABC is an informant-based rating instrument that was designed to measure several maladaptive behaviors. There are a total of 58 items, each item scored on a 4-point scale (0: not a problem at all, through 3: problem behavior is present to a severe degree). There are a total of five subscales: (1) *Irritability, Agitation*, (2) *Lethargy, Social Withdrawal*, (3) *Stereotypic Behavior*, (4) *Hyperactivity, Non-Compliance*, and (5) *Inappropriate Speech*. For the purposes of the current study, the *Hyperactivity* subscale, which consists of 15 items, was used as a potential predictor of RRB phenotypic cluster membership.

Child Behavior Checklist (CBCL; Achenbach, & Rescorla, 2001). The CBCL is a parent-report measure comprised of 120 questions that assess internalizing symptomology related to psychopathologies such as anxiety or mood disorders, and externalizing behaviors relating to behavioral disorders (e.g., attention-deficit/hyperactivity disorder, oppositional defiant disorder). There are a total of 8 syndrome scales, which include: *Withdrawn/Depressed, Somatic Complaints, Social Problems, Rule Breaking Behavior, Anxious/Depressed, Attention Problems, Aggressive Behavior*, and *Thought Problems*. The *anxiety* problems subscale T-score was used in this study.

The Repetitive Behaviors Scale- Revised (RBS-R; Bodfish, et al., 2000) is a parent questionnaire that exclusively measures the type and severity of RRBs exhibited by individuals with ASD. The measure is comprised of 43 questions grouped into 6 conceptually derived

21

subscales, which include: *Stereotyped Behavior, Self-injurious Behavior, Compulsive Behavior, Ritualistic Behavior, Sameness Behavior*, and *Restricted Behavior* (See Appendix 1). All of 43 RBS-R items were included in the factor analyses to determine the underlying factor structure, which resulted in the new RBS-R factor scores. Once the new factors were established, factor sum scores were converted into standardized z-scores with a mean value of zero and standard deviation of one for all subsequent analyses.

Vineland Adaptive Behavior Scale- Second Edition (VABS-II; Sparrow, Cicchetti, & Balla, 2005). The VABS is a clinician-administered, semi-structured parent interview designed to measure adaptive behavior skills across three domains: *Communication, Socialization* and *Daily Living Skills*. The VABS-II provides an indicator of the degree to which adaptive skills are impacted in children with developmental disabilities. Under the subcategory of *Socialization,* there is a subset of questions labeled *Coping Skills*, which includes items such as "Changes easily from one at-home activity to another" and "Changes behavior depending on how well he or she knows the other person".

Statistical Analyses

Descriptive information was calculated for the entire sample of probands as well as descriptive statistics for individual datasets. Due to the unique inclusion criteria in the SSC sample, differences between SSC participants and Simons Ancillary participants were examined to detect the possible presence of significant differences in basic demographic information as well as their reported RRBs. No significant differences existed between the data sets, indicating acceptability in combining datasets to explore the current study's aims.

Exploratory Factor Analysis (EFA)

The first aim of this study was to examine the factor structure of the Repetitive Behavior Scale- Revised (RBS-R) to determine how many unique RRB forms are measured. The first step in determining the factor structure was to run an exploratory factor analysis of the 43 RBS-R items using Mplus Version 7 (Muthen & Muthen, 2012), an oblique CF-quartimax rotation (Gorsuch, 1983) and a weighted least- squares with mean and variance adjustment (WLSMV) to account for the ordinal nature of the data (Muthen, DuToit, & Spisic, 1997). EFA assumes that each variable, in this case each question on the RBS-R, may be associated with any other factor without an a priori hypothesis about factors or variables (Finch & West, 1997).

To determine the optimal number of factors, a combination of model fit statistics and examination of factor loadings were used. The chi-square value is typically an informative model fit statistic; however, the chi-square test is sensitive to sample size, such that large samples often result in statistically significant chi-square values (Satorra & Bentler, 2001). Given the number of participants in the current dataset, the chi-square values were analyzed with caution. Additional model fit statistics included root mean square error of approximation (RMSEA; Marsh, Balla & Hau, 1996), the Standardized Root Mean Square Residual (SRMR; Hu & Bentler, 1999), the Comparative Fit Index (CFI; Bentler, 1990) and the Tucker Lewis Index (TLI; Bentler, 1990). The RMSEA is a measure of model fit that is not as sensitive to sample size and values below .06 indicate an acceptable model fit (Satorra & Bentler, 2001). The SRMR is another descriptive model fit statistic in which lower values indicate better model fit, with a suggested cut-off of .08 or below (Hu & Bentler, 1999). Lastly, both the CFI and TLI are typically presented together in EFAs and both serve as measures of model fit, ranging from 0 to 1 with higher values indicating better fit and cutoff scores of .90 (Hu & Bentler, 1999).

Determining Factor Structure. As items were permitted to load on only one factor for the CFA, items that loaded significantly >.30 on more than one factor were evaluated to determine the ideal factor pattern. Individual items, or in the case of the current study, individual questions from the RBS-R, were independently assessed for conceptual fit on the factor they most strongly loaded on to determine if it is an appropriate factor fit considering the other items that loaded strongly on the respective factor. The overall goal of EFA was to identify factors, based on a given dataset, and maximize the amount of variance explained by the model (Suhr, 2006). Once a model has been theoretically and/or statistically established and hypotheses have been made, a confirmatory factor analysis can inform the likelihood of the hypothesized results.

Confirmatory Factor Analysis (CFA)

A Confirmatory Factor Analysis (CFA) was conducted once the relationships among variables were established through statistical analyses and a theoretical model was evaluated (Suhr, 2006; Thompson, 2004). While the EFA allows for all items to load on any factor, the CFA restricts the factors on which items load. Each item was permitted to load on only one factor. Model fit was determined using recommended indices of model fit including Chi-Squared test, RMSEA, RMR, CFI and TLI. Additionally, the CFA model produced a weighted root mean square residual (WRMR) that is an empirically supported measure of model fit comparable to the other fit indices and is suggested to be highly useful for data that isn't normally distributed (Muthen & Muthen, 2001; Yu, 2002; Hu & Bentler, 1999). A WRMR value above 1.0 is considered good model fit. Factor loadings from the CFA were reported as the standardized model estimate loadings and associated standard errors.

K-Means Cluster Analysis

Cluster analysis provides a unique approach to examining which results in the identification of patterns that organize variables into taxonomies, grouping cases with similar patterns together (Lloyd, 1982). For the current study, the K-means cluster analysis was run to systematically and conceptually group participants with similar RRB patterns together.

The newly established factors from the CFA of the RBS-R were used to examine the various patterns of RRB presentation for this population. The goal of a k-means clustering is to partition individuals into clusters where every participant belongs to a cluster with others presenting with similar patterns (Hartigan & Wong, 1979). The optimal number of clusters must strike a balance between successfully compressing the data as a single cluster would, while maintaining maximum accuracy where every participant is assigned to its own cluster. The optimal number of clusters for the data was determined using both theoretical and empirical considerations. Previous research exploring RRBs have defined between two and six distinct *types* of RRBs; yet, there hasn't been a clustering of those RRBs into distinct profiles to serve as a comparison or as an empirical rationale to test the fit of a specific number of clusters. Therefore, comparisons of three, four, five and six cluster solutions were conducted.

One approach that was used to determine model fit for each cluster was to examine the number of iterations it took to satisfy the convergence criterion (i.e., reach 0.00 (Inaba, Katoh, & Imai, 1994). There is no guarantee that data will cluster and iterate to convergence quickly, if at all. Therefore this is a reasonable justification for this approach in determining the fit between the number of clusters and the data being analyzed. Statisticians have concluded that it is acceptable to institute a maximum criterion of between 15 and 20 iterations for the data to reach

convergence criterion where the clusters optimally fit the data. Cluster statistics were explored after running three, four, five and six cluster solutions; results are described below.

Multinomial Logistic Regression

The final research aim was to determine the ability of several behavioral and developmental characteristics to predict cluster membership. Correlation analyses among all predictors were conducted prior to running the MLR to determine presence of collinearity. A multinomial logistic regression was run with individual cluster assignment as the outcome variable and participants' standardized scores of ASD severity, nonverbal IQ, hyperactivity, anxiety, and coping skills as predictors. Age differences across clusters was independently examined by running a one-way ANOVA prior to running the MLR to determine if age significantly differed among clusters.

The MLR provides a unique approach to determine the odds ratio of an individual being in one cluster relative to the odds of them being in the comparison cluster based on several characteristics (i.e., predictors). Therefore, it is important to choose a comparison cluster that will provide the most robust information in the analysis of these comparison solutions. Prior to exploring the individual cluster phenotypes to decide on a comparison cluster, the options were carefully considered and a conceptual decision was made. The comparison group should be the one that differs the most from the others, or the group that could be considered the "optimal outcome" group that possesses characteristics that researchers would want to test and discover what makes that group of participants different (Fields, 2009). Therefore, the cluster with the lowest levels across all RRBs was used as the baseline comparison cluster.

Goodness of fit of the MLR model was assessed using the log-likelihood (LL), which sums the probabilities of predicted outcomes and actual outcomes, analogous to the residual sum

of squares in typical multiple regression. That is, the LL variable indicates how much unexplained data remains after the model is fit; where large values of the LL statistic tends to describe a poor fit for the model (Field, 2009). Results of the multinomial logistic regression produced significance statistical values, which indicated the extent to which individual characteristics were able to significantly predict membership to one cluster over another.

The individual parameter estimates for each comparison between the optimal profile group vs. the other profiles were individually examined to determine the significant and non significant results across predictor variables and interactions. The significance values were used to determine which of the characteristics were significant in predicting profile membership, with the odds ratio statistic indicating the odds of a participant being in a cluster when compared to the odds of them being a member of the optimal outcome profile group. Overall model fit statistics as well as individual parameter estimates of the multinomial logistic regression were examined.

RESULTS

Descriptive Statistics

2,856 participants ranged in age from 4 years old to 18 years old, with a mean age of 9 years old (108.31 mos.; *SD*=42.81). The majority of participants were male (83.4%) and Caucasian (78.5%), with 4% Black or African American, 4% Asian, 7.8% responded as more than one race, .1% Native Hawaiian, .2% Native American, and 4.5 % were categorized as "other". Additionally, 11.2% of all participants considered themselves Hispanic or Latino/a. The sample was also highly educated, with 87.2% of mothers and 81.7% of fathers having *at least* some college education, indicating a much higher socioeconomic status of SSC participants

compared to the general population. More than half of all participants (*n*=1566, 54.9%) completed Module 3 ADOS and the average calibrated severity score (CSS) from the ADOS was 7.4 (see Tables 1.1 and 1.2 for full demographic information).

The Five-Factor Solution of the RBS-R

Results from the exploratory factor model revealed that the 5-factor solution indicated the best model fit. Fit indices are as follows: χ^2 (698) = 3852.53, *p* < .001; CFI= .952, TLI= .938, RMSEA = .04; SRMR= .036. The values for each model fit index indicated a good fit to the data, with both CFI and TLI values meeting the recommendation of above the .9 cut-off and close to a value of 1 (Bentler, 1990), SRMR falling well below the < .08 cut off, and RMSEA of .04 falls below .06. Refer to Table 2 for both EFA and CFA model fit indices as well as the recommended range for each fit statistic.

Table 1.1.
Participant demographic information

	N	%
Males	2382	83.4
Females	375	13.1
Missing	98	3.4
Race		
White	2167	78.5
African American	111	4.0
Asian	111	4.0
More than one race	215	7.8
Native American	6	.2
Native Hawaiian	2	.1
Not specified/Other	147	5.3
Ethnicity		
Hispanic	308	11.2
Non-Hispanic	2445	88.6
ADOS Module		
1	514	18.0
2	603	21.1
3	1566	54.9
4	74	2.6
Missing	98	3.4

Table 1.2.

Participant Descriptive Characteristics

Variable	M	SD	Range
Age	108.3	42.8	(48, 216)
NVIQ	84.5	26.3	(9, 161)
VIQ	78.1	31.4	(5, 167)
CSS	7.4	1.7	(4, 10)
ADOS RRB	4.0	2.0	(0, 8)

Table 2.

Model fit indices optimal values and fit statistics from EFA and CFA

Goodness of fit Statistic	Symbol	Optimal Range	EFA Results	CFA Results
Chi-square test	χ^2	$P < .05$	4565.3*	4589.7*
Root Mean Square Error of Approximation	RMSEA	$< .06$	.04	.053
Tucker-Lewis Index	TLI	$> .6$	.938	.916
Comparative Fit Index	CFI	$>.6$	.952	.923
Standard RMR	SRMR	$<.08$	.036	N/A
WRMR	WRMR	>1	N/A	2.492

Note. * = $p < .001$

Individual RBS-R items and corresponding factor loadings were examined to ensure that each factor that was derived from the EFA had strong cohesion among items and were both statistically and conceptually strong. There were nine items on the RBS-R that did not significantly load on a single primary factor (λ=-0.16 - 0.28), or had low loadings on more than one factor (λ=0.29 – 0.31) and were removed from the model (see Appendix B for the entire model factor loadings). RBS-R item numbers 20 – 27 (the original *Ritualistic* subscale) were all removed from the final factor model after examination of their loading pattern and conceptual fit with remaining factors. The remaining 34 RBS-R items successfully loaded onto five distinct factors that displayed strong statistical and intuitive cohesion. The new factors were used in all subsequent analyses; the five factors were specified as *hand/body mannerisms, self-injurious*

behaviors, compulsive behaviors, perseverative interests, and rigidity. Specific examples can be found below in Table 3.

Table 3.

The five-factor result from the EFA with brief examples

Factor Name	Example Behaviors
Hand/Body	*Flapping, toe walking, body rocking, spinning objects turning, jumping*
Self Injurious	*Biting, hitting, scratching, banging, pulling, picking, etc. of self*
Pers. Interests	*Fascination, preoccupation with subject/activity, attached to object/s*
Compulsiveness	*Arranging objects, counting, checking, hoarding, washing/cleaning*
Rigidity	*Inflexibility, resisting change, insistence on sameness*

Note. Rigidity is made up of most of the original "sameness behavior" RBS-R subscale

Confirmation of the Five-Factor Solution of the RBS-R

The specified CFA model included the 34 selected items from the RBS-R. The CFA model fit indices were: $\chi^2 (517) = 4589.7$, $p < .001$, CFI= .923, TLI= .916, RMSEA= .053 and WRMR= 2.492. The values for each model fit index indicated a good fit to the data, with both CFI and TLI values meeting the recommendation of above the .9 cut-off and close to a value of 1 (Bentler, 1990), RMSEA fell just below .06, and the WRMR result of 2.492 surpassed the threshold of 1, indicating good model fit (Muthen & Muthen, 2001). Each factor was then calculated by summing the item scores, resulting in the following mean factor scores: Factor 1 Hand/Body ($M = 5.06$), Factor 2 Self- Injury ($M = 2.09$), Factor 3 Compulsivity ($M = 5.56$), Factor 4 Rigidity ($M = 7.11$), Factor 5 Perseverative Interests ($M = 2.44$).

Phenotypic Clusters Characteristics

Standardized factor scores (i.e., z-scores) from the CFA were used for the k-means cluster and all subsequent analyses. The five-cluster solution was derived after comparisons were made across three, four, five and six cluster solutions. The three-cluster solution resulted in high,

low and mixed phenotype clusters, failing to converge after 20 iterations. Similar results were found for the four and six cluster solutions; which also failed to converge to 0.00 after 20 iterations. However, the five-cluster solution successfully converged to 0.00 across all cluster centers in 20 iterations (see Appendix A for cluster iteration history). Basic descriptive characteristics for the five-cluster solution can be found in Table 4.

Figure 2. Final five cluster solution of standardized RRB factor sum z-scores
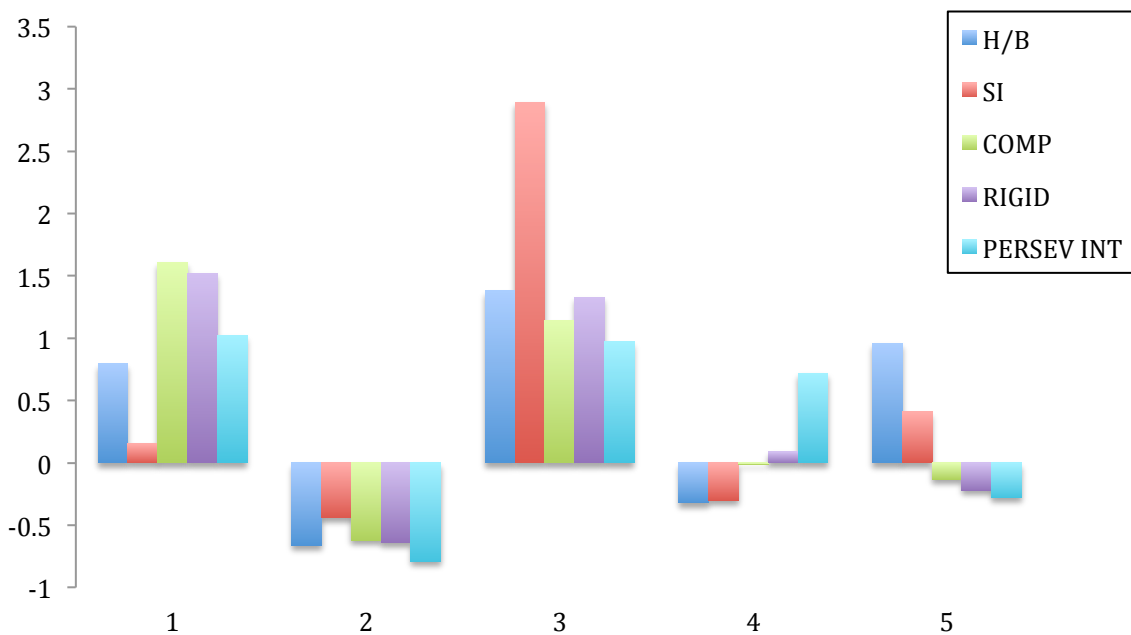
Note. Z-scores have a mean of 0 and a standard deviation of 1.

The two most distinct clusters were the all *high* and all *low* across all RRB subtypes, which included the most participants (Cluster 2 "All Low"; $n = 1138$) and the lowest amount of participants (Cluster 3 "All High"; $n = 154$). Cluster 1 "High Except SI" presents comparable levels of compulsivity, rigidity and perseverative interests to the "All High" cluster, with considerably higher self-injurious behaviors exhibited. Cluster 5 is labeled "Classic Low Order" due to its pattern of minimal levels of compulsivity, rigidity or perseverative interests, yet

presents hand/body and self injurious behaviors above the mean, which represents the phenotype

of individuals presenting the low order RRBs described previously. Cluster 4 "Mixed-High

Perseveration" presents nearly the inverse of Cluster 5, with low scores of hand/body

mannerisms and self-injury; yet the highest RRB presented in this profile group is perseverative

interests.

Table 4.
Descriptive characteristics of the five-cluster solution with labels

Cluster	Label	*n*	Age μ (SD)	Range	IQ μ (SD)	Range
1	"High Except SI"	372	106.5 (41.4)	(49, 215)	80.1 (29.6)	(14, 161)
2	"All Low" *	1138	110.2 (45.2)	(48, 216)	87.8 (29.5)	(13, 254)
3	"All High"	154	107.3 (40.6)	(49, 215)	70.6 (33.5)	(11, 182)
4	"Mixed- High Perseveration"	700	110.7 (41.5)	(48, 214)	87.8 (29.2)	(12, 204)
5	"Classic Low Order"	491	102.2 (40.2)	(48, 211)	64.74 (31.9)	(7, 158)

An independent samples *t*-test revealed that gender was not significantly different

between the clusters (t (2755) = 1.39, p = .166) after the assumptions of homogeneity of

variances were tested and satisfied via Levene's F test; F (2755) = 3.18, p = .075. Next, a one-

way ANOVA was conducted to examine whether there were statistically significant age

differences across the five clusters. Results revealed statistically significant differences among

the clusters, F (4, 2752) = 3.66, p = .006. Post-hoc Games-Howell tests revealed Cluster 5 (μ =

102.2, SD = 40.2) significantly differed from both Cluster 2 (μ = 110.2, SD = 45.2) and Cluster 4

(μ = 110.7, SD = 41.5). There were no other significant differences between the other groups.

Final cluster standardized factor scores are found in Table 5 and depicted visually in

Figure 2. Basic descriptive information was calculated for the clusters, which included age, IQ as

well as several behavioral descriptive characteristics that went into the MLR model. Initial

inspection of basic cluster characteristics revealed that close to half (40%) of participants were in

the "All Low" cluster (n = 1138), and the smallest group were the "All High" cluster made up

only 5% of the total group (n = 154). All five clusters contained a similar age range of

participants (49 mos. to 216 mos.).

Table 5.
Average standardized factor sum z-scores by cluster membership

	Clusters				
	1	2	3	4	5
Hand/Body	.79462	-.66092	1.37919	-.32308	.95781
Self-Injury	.15569	-.43692	2.88603	-.29860	.41520
Compulsivity	1.61329	-.61998	1.14489	-.00675	-.13483
Rigidity	1.52543	-.63649	1.32784	.08641	-.22017
Perseverative Interests	1.01566	-.78560	.96609	.72109	-.27973

Note. Z-scores have a mean of 0 and a standard deviation of 1.

Predictive Power of Individual Characteristics

Cluster membership was entered as the dependent variable in the Multinomial Logistic

Regression (MLR) to determine the predictive power of several behavioral and developmental

characteristics in determining cluster membership. The predictors entered into the model

included the standardized scores of hyperactivity, anxiety, coping skills, nonverbal IQ, and

ADOS calibrated severity scores (CSS). Cluster 2, the *"All Low"* profile had the most unique

RRB profile compared to the other clusters, as well as being the ideal group with the lowest RRB

scores overall; which would provide the most robust findings when used as the comparison

cluster in the MLR analysis. Table 6

Table 6.

Average standardized scores of characteristics for the five RRB profiles

	Cluster 1 n= 372	Cluster 2 n= 1138	Cluster 3 n= 154	Cluster 4 n= 700	Cluster 5 n= 491
Age (mos.)	106.5	110.2	107.3	110.7	102.2
NVIQ	80.1	87.81	74.6	87.8	67.74
Hyperactivity	22.3	11.3	28.3	16.8	20.43
CSS	7.6	7.3	7.4	7.4	7.7
Anxiety	65.35	57.8	66.45	61.41	60.31
Coping	9.6	10.9	8.95	10.4	9.5

Note. NVIQ= Nonverbal IQ ratio score, *Hyperactivity=*domain T-score from the ABC, *CSS=* Calibrated severity score from the ADOS, *Anxiety=* domain T-score from the ABC, *Coping=* domain T-score from the Vineland.

A forward entry stepwise multinomial logistic regression model was run with the most significant variables added one at a time. The process of adding variables that had the greatest impact on the model continued until none of the remaining variables would significantly contribute to the model. The first variable entered into the model with the most significant influence on determining cluster membership was *anxiety* (χ^2 (4) = 156.80, *p* < .001), followed by *hyperactivity* (χ^2 (4) = 698.47, *p* < .001), and finally *coping skills* (χ^2 (4) = 80.82, *p* < .001). As seen in Table 7, after the predictor variables were entered into the model, the AIC and BIC values decrease, indicating that the fit of the model was significantly improved. Both *nonverbal IQ* and *ADOS calibrated severity scores (CSS)* were omitted from the model, as they failed to significantly contribute to the model over and above the included predictors.

Model Fit. MLR models create several informative values to determine significance of the model (i.e., AIC, BIC, Log Likelihood) and of the model fit to the data (i.e., Goodness of Fit Pearson and deviance statistics). First, to test the model itself, the log-likelihood (LL) measured

how much unexplained variability there was in the data. The change in LL indicates how much

new variance was explained through the model by testing the decrease in variance between the

baseline model ($log \lambda = 7909.32$) and the final model ($log \lambda = 6973.24$). The chi-square test of

differences between variances resulted in a significant model fit χ^2 (12) $= 936.01$, $p < .001$

indicating that the final model explains a significant amount of the original variability.

Table 7.
Model Fitting Information

Model	Model Fitting Criteria			Likelihood Ratio Tests		
	AIC	BIC	-2 Log Likelihood	Chi-Square	df	Sig.
Intercept Only	7917.325	7940.996	7909.325			
Final	7005.239	7099.925	6973.239	936.086	12	.000

The Goodness of fit statistics indicate how well the model fits the data, which answers

whether the predicted values from the model differ significantly from the actual observed values

(Fields, 2009). Both the Pearson statistic ($p = .81$) and the deviance statistic ($p = 1.00$) were not

significant, indicating the model had strong predictive power and fit the data. The two measures

of R^2, Cox and Snell ($R^2 = .289$) and Nagelkerke ($R^2 = .306$) also concluded that the model was

of significance (Cox & Snell, 1989; Nagelkerke, 1991).

Table 8.1.
Goodness-of-Fit

	Chi-Square	df	Sig.
Pearson	10832.780	10964	.812
Deviance	6973.239	10964	1.000

Table 8.2.
Pseudo R-Square

Cox and Snell	.289
Nagelkerke	.306

Likelihood ratio tests indicated that anxiety (χ^2 (4) = 153.16, $p < .001$), hyperactivity (χ^2 (4) = 475.51, $p < .001$) and coping (χ^2 (4) = 80.82, $p < .001$) were all significant predictors of the model. However, in order to determine what the specific effects were, the parameter estimates were examined.

Individual Parameter Estimates. MLR analyzes outcomes dyadically, necessitating the effect statistics to be interpreted individually. Of the variables included as potential predictors in the MLR model, *anxiety* was the strongest predictor of a participant's cluster membership in comparison to Cluster 2 (our predetermined comparison cluster of "All Low" RRBs). The odds ratio Exp(*B*) indicates the change in odds of predicting whether a participant was a member of a particular cluster over being a member of Cluster 2. The strongest odds ratio in the model was in predicting membership to Cluster 3 ("All High") compared to Cluster 2 (*b* = .17, Wald χ^2 (1) = 247.55, $p < .001$) which indicates that as a participants' score of *hyperactivity* increases by one, the odds of them belonging to Cluster 3 (vs. Cluster 2) increases by 1.2. *Anxiety* also significant in increasing a participants' chance of being in Clusters 1, 3, 4 or 5 compared to Cluster 2 with each comparison exhibiting significant contribution of anxiety scores (*p* < .001) to the model and Exp(*B*) values ranging from 1.02 to 1.09. Lastly, *coping* also significantly contributing to predicting membership, each of the odds ratios are less than one indicating *less* of a chance that they would belong to a given cluster over the comparison Cluster 2 (see Table 10 for full parameter estimates). Most notably, both *non-verbal IQ* and *calibrated severity scores (CSS)* of ASD symptoms were not significant contributors to the model, and thus the MLR automatically excluded those variables from the analyses, as they failed to significantly contribute to the prediction model.

Table 9.

Final reduced model fit statistics

	Model Fitting Criteria			Likelihood Ratio Tests		
	AIC of Reduced Model	BIC of Reduced Model	-2 Log Likelihood of Reduced Model	Chi-Square	df	Sig.
Effect						
Intercept	7174.399	7245.414	7150.399	177.160	4	.000
Anxiety	7150.396	7221.411	7126.396	153.157	4	.000
Hyperactivity	7472.753	7543.768	7448.753	475.514	4	.000
Coping	7078.055	7149.070	7054.055	80.816	4	.000

Note. The chi-square statistic is the difference in -2 log-likelihoods between the final model and a reduced model.

Table 10.

Multinomial Logistic Regression parameter estimates

		95% CI for Odds Ratio		
	b (SE)	Lower	Odds Ratio	Upper
1 vs. 2				
Anxiety	-.079 (.008) ***	1.066	1.082	1.098
Hyperactivity	.111 (.007) ***	1.101	1.117	1.134
Coping	-.157 (.031)***	.804	.855	.908
3 vs. 2				
Anxiety	.086 (.011) ***	1.068	1.090	1.113
Hyperactivity	.166 (.011) ***	1.156	1.180	1.205
Coping	-.284 (.049)***	.684	.753	.828
4 vs. 2				
Anxiety	.041 (.006) ***	1.030	1.042	1.054
Hyperactivity	.064 (.006) ***	1.053	1.066	1.078
Coping	-.046 (.022)*	.914	.955	.998
5 vs. 2				
Anxiety	.022 (.007)***	1.009	1.023	1.037
Hyperactivity	.096 (.007)***	1.086	1.101	1.115
Coping	-.195 (.027)***	.780	.823	.868

Note. R^2= .29 (Cox & Snell), .31(Nagelkerke). Model x^2(12)= 936.01, p < .00. (* p < .05, ** p < .01, *** p < .001).

DISCUSSION

The recent changes to the DSM have created a more comprehensive list of RRB subtypes than were previously included and set a more stringent benchmark to meet criteria in the RRB domain. Such changes reflect the progression of research supporting the importance and independence of RRBs as an integral component of diagnosis, rather than a by-product of the "core" social communication impairments (Richler, et al., 2010; Kanne, 2013). From its earliest conception, ASD has been characterized by the presence of frequently and highly repetitious behaviors, with a marked desire for environmental sameness and consistency (Kanner, 1943). Yet, this complex behavioral domain is historically under-represented in research efforts and falls secondary to social communication deficits in ASD research. Reviews of past studies on RRB presentation have highlighted issues including a lack of methodological consistency, with varying approaches to defining, organizing, and measuring RRBs. These discrepancies have led to splintered advancements in understanding the etiology, early behavioral manifestations and longitudinal developmental implications of RRBs (Leekam, 2011).

The primary aims of this study were to characterize RRB phenotypes of individuals with ASD and to determine the influence of developmental and behavioral characteristics on RRB profiles. This study revealed that there were five distinct RRB subtypes captured by the RBS-R, with five distinct phenotypic profiles generated from those subtypes. Hyperactivity, anxiety and coping skills significantly predicted participants' RRB phenotype, while IQ and symptom severity had little effect. The findings in this study provide a unique perspective when conceptualizing ASD symptomology and the influence of non-ASD specific traits on this core domain.

The Five- Factor Model of RRB Types

The five-factor model result from the factor analyses of the RBS-R exhibits substantial consistency with previous studies examining the factor structure of the RBS-R (Bishop, et al., 2013; Lam & Aman, 2007). Comparisons between factor results of the RBS-R can be seen in Figure 3. Most notably, the current study excluded 3 items from the original compulsive scale (items included hoarding, repeating events, and touching/tapping items repeatedly) as the item factor loadings were above .4 on more than two newly calculated factors. These results indicated that there wasn't a single factor that accounted for the variability of each item, forcing those items to be excluded. Similarly, five items on the original ritualistic scale were excluded which included items regarding eating, sleeping, travel, play and self-care as they were highly loaded (ranging from .5-.7) on multiple factors. These findings indicate they may not be sufficiently differentiating types of RRBs measured by each question, which leads investigators to wonder if the questions are adequately differentiating between RRB subtypes.

Bishop, et al. (2013) investigated RRB data from both the ADI-R and the RBS-R and found that the ADI-R items resulted in a two-factor (RSM and IS) model, whereas the RBS-R resulted in a five-factor model as the best fit. When examined in conjunction with findings from the Lam & Aman (2007) study as well as the current results, it is evident that using a measure with a wider range of questions such as the RBS-R provides more in-depth and informative results when examining the specific types and severities of RRBs.
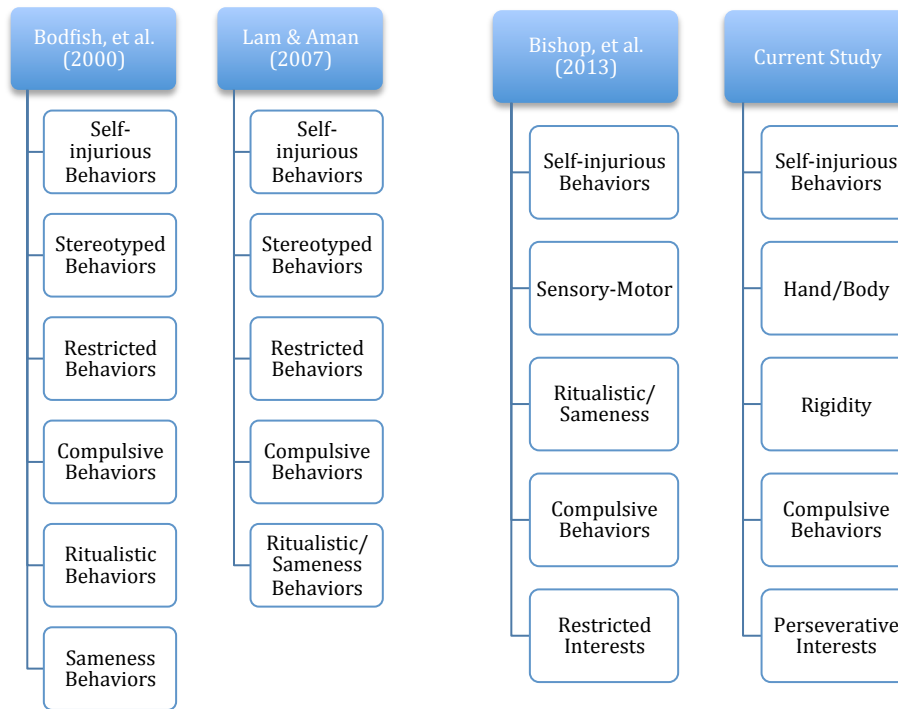
Despite the utility of a measure dedicated to specific RRB types and severity, factor results from previous studies fail to be substantiated with each study, leading to the conclusion that a final set of RRB subtypes (factors) have yet to be established unequivocally across studies. Further, each analytic result has not been entirely consistent with the six conceptually derived

39

subscales that Bodfish, et al. (2000) originally established. Discrepancies between the subtypes and the original subscales, as well as between the previously proposed models can be seen below in Figure 3. As previously discussed, RRBs comprise a complex and heterogeneous set of behaviors that vary greatly depending on the population being measured; therefore, it is not a complete surprise that each factor analytic study has resulted in slightly altered structures. However, given the vast age range include in the current study and largest number of participants to date for an RBS-R factor analysis, the resulting factor structure warrants consideration as an organizational RRB factor structure to be analyzed for confirmatory analyses in future studies using the RBS-R.

The RRB subtype found most consistently across studies has been self-injurious behaviors. As seen in Figure 3, researchers who organized and defined more than two categories of RRBs had one striking consistency, the inclusion of an independent category of self-injurious behavior (Lam & Aman, 2007; Bodfish, et al., 2000; Mirenda, et al., 2010; Bishop, et al., 2013).

Further, self- injury is arguably the most recognizable and disruptive RRB consistently found to be related to greater impairment with significantly lower IQ and higher severity of ASD symptoms (Bodfish, et al., 2000; Bishop, Richler, & Lord, 2006). In fact, the most recent study examining RRB subtypes concluded that SI behaviors create significant difficulty in dichotomizing RRBs, as the SI items fail to load with the repetitive sensory motor category *or* with the insistence on sameness supporting the existence of additional subcategories (Bishop, et al., 2013). Further, SI is the only RRB subtype to consistently load *identically* as an entire subscale in every factor analytic study of the RBS-R, which was also true in the current study, indicating its distinctiveness (Bodfish, et a., 2000; Lam & Aman, 2007; Bishop, et al., 2013).

Figure 3. Results of previous RBS-R factor analyses

The RRB subtype found most consistently across studies has been self-injurious behaviors. As seen in Figure 3, researchers who organized and defined more than two categories of RRBs had one striking consistency, the inclusion of an independent category of self-injurious behavior (Lam & Aman, 2007; Bodfish, et al., 2000; Mirenda, et al., 2010; Bishop, et al., 2013). Further, self- injury is arguably the most recognizable and disruptive RRB consistently found to be related to greater impairment with significantly lower IQ and higher severity of ASD symptoms (Bodfish, et al., 2000; Bishop, Richler, & Lord, 2006). In fact, the most recent study examining RRB subtypes concluded that SI behaviors create significant difficulty in dichotomizing RRBs, as the SI items fail to load with the repetitive sensory motor category *or* with the insistence on sameness supporting the existence of additional subcategories (Bishop, et al., 2013). Further, SI is the only RRB subtype to consistently load *identically* as an entire

subscale in every factor analytic study of the RBS-R, which was also true in the current study, indicating its distinctiveness (Bodfish, et a., 2000; Lam & Aman, 2007; Bishop, et al., 2013).

Phenotypic Clusters of RRBs

Cluster analysis provides a novel approach to statistically explore phenotypic profiles and the co-occurrence of RRB types and severity across individuals with ASD. This is the first study of its kind to statistically generate clustered phenotypes, each consisting of multiple RRB subtypes. RRBs don't occur in isolation; the pattern of behavior is fluid with minimal evidence to explain the variations seen across and within individuals. By studying RRBs in a way that allows for multiple RRBs to co-occur at varying levels, researchers may gain a more accurate and informative picture of how these behaviors manifest across individuals with ASD. However, when researchers rely solely on parent report measures, there is a limited scope of distinct behaviors from which combination or cluster phenotypes can be derived. Therefore, it is recommended that multiple modalities be used to capture and characterize RRBs to gain a better understanding of how they evolve over time, across contexts and as a result of individual characteristics.

Predictive Characteristics of RRB Profiles

Historically, the two most universally agreed upon characteristics found to significantly relate to RRB presentation have been age and IQ (Esbensen, et al., 2009; Georgiades, et al., 2010; Mirenda, et al., 2010; Richler, et al., 2010). It has even been noted that due to methodological limitations and specific study aims, "untangling relationships between (RRB) subscale scores and age and IQ has not been possible in the majority of RBS-R studies to date" (Bishop, et al., 2013). Taking a novel approach to address this issue in this study has helped to clarify some of the previous findings, and subsequent theories, regarding the relationship

between age, IQ and RRBs. The basic correlational relationships between RRBs, age, and IQ have been replicated across studies; however, a basic correlation analysis is a missed opportunity to ask the more informative research question of how they are related. In other words, instead of addressing *IF* RRBs, age, and IQ are related to one another, it should be *HOW* are they related. More specifically, how much does one variable account for or predict the others. Surprisingly, as the current study showed, although IQ may be an important predictor of the presence of *any* RRBs, it is not the most important factor in determining a specific RRB phenotype.

Lastly, it should be noted that *coping* was significant in the MLR model, yet with a very distinct influence on cluster prediction in comparison to anxiety and hyperactivity. That is, each of the odds ratios were less than one indicating *less* of a chance that they would belong to any of the clusters in comparison to Cluster 2 ("All Low"). This finding is consistent with our understanding of maladaptive behavior and RRBs, as an increase in coping behaviors would mean better adaptive skills such as managing emotionally arousing situations, and exerting self-regulatory control, thus reducing likelihood of engaging in RRBs.

The association between phenotypic patterns of RRB expression in ASD and the behavioral proxies for common comorbid symptomology in ASD has not yet been clearly defined. However, given the results of Rao & Landa (2014) on the additive effect of presenting behavioral phenotypes spanning across both diagnostic categories of ADHD *and* ASD on the severity of RRB presentation, it is not surprising that *hyperactivity* was the strongest predictor of RRB cluster membership. Further, the finding that non-ASD specific traits were able to significantly predict variations in half of the diagnostic dyad of behaviors warrants continued investigation.

Limitations

Results from the current study should be interpreted and applied somewhat cautiously for several reasons. First, this study examined data from a racially homogeneous sample, which is not reflective of the general population. Future studies should include a more diverse sample with more variety in the economic status of participating families as well as a more accurate sampling and balanced representation of racial groups. Finally, the sample size included in this study is the largest to date compared to similar studies. However, without analyzing longitudinal data, it's impossible to statistically or methodologically conclude how RRBs will change overtime within individuals, which prohibits the current study to be able to conclude how these phenotypic profiles change overtime within individuals. Specifically, relationships described by this study's findings may also shift over time along with the relationship between RRBs and influence those shifts will have on other developmental and clinical characteristics for individuals with ASD.

Conclusion

Repetitive behaviors can present significant barriers for individuals with ASD and their families. Therefore, it is important to continue to examine the ways in which this set of heterogeneous behaviors relate to specific developmental or behavioral characteristics; particularly those which have not been examined in previous studies. Results find that symptoms of hyperactivity and anxiety are better predictors of RRB subtypes than are IQ and age. Moreover, coping strategies are also important in the expression of RRBs. The ultimate goal in this field of research will be to understand the function that RRBs serve, related traits, and finally to use this information to inform effective intervention strategies. Therefore, researchers should continue to explore RRB phenotypic profiles and identify related characteristics to continue to

discover the interaction effects of individual traits and RRB presentation This study provides a

significant foundation in understanding the complex nature of this diverse behavioral phenotype.

Appendix A.

EFA 5-factor iteration history reaching convergence in 20 iterations to support model fit

Iteration	Change in Cluster Centers				
	1	2	3	4	5
1	2.408	2.773	2.631	2.581	2.586
2	.756	.438	.666	.326	.280
3	.278	.108	.321	.120	.158
4	.135	.045	.228	.079	.107
5	.075	.035	.147	.066	.108
6	.066	.028	.143	.052	.119
7	.068	.023	.078	.049	.095
8	.052	.027	.066	.044	.088
9	.054	.013	.050	.032	.057
10	.045	.014	.027	.027	.033
11	.037	.010	.037	.019	.033
12	.018	.010	.041	.024	.028
13	.026	.006	.025	.025	.021
14	.024	.006	.036	.020	.020
15	.010	.006	.017	.014	.018
16	.009	.005	.016	.010	.010
17	.008	.005	.020	.007	.013
18	.010	.004	.014	.010	.008
19	.004	.001	.000	.002	.003
20	.000	.000	.000	.000	.000

a. Convergence achieved due to no or small change in cluster centers. The maximum absolute coordinate change for any center is .000. The current iteration is 20. The minimum distance between initial centers is 5.331.

Appendix B. EFA final factor loadings for the five-factor solution

	Factor 1	Factor 2	Factor 3	Factor 4	Factor 5
RBS1	0.136*	-0.032	0.539*	0.162*	-0.177*
RBS2	0.150*	0.016	0.506*	0.101*	-0.104*
RBS3	0.083*	-0.011	0.595*	0.028	-0.102*
RBS4	0.182*	-0.013	0.577*	0.048*	-0.053
RBS5	0.173*	0.047	0.564*	-0.028	0.086*
RBS6	0.209*	0.079*	0.445*	0.035	0.102*
RBS7	0.743*	-0.071*	0.099*	0.066*	-0.080*
RBS8	0.719*	-0.046	0.115*	0.092*	-0.028
RBS9	0.812*	-0.075*	0.051	0.077*	0.047*
RBS10	0.506*	0.018	0.085*	-0.006	-0.04
RBS11	0.654*	0.088*	-0.034	-0.058	0.01
RBS12	0.643*	0.195*	-0.114*	-0.100*	0.044
RBS13	0.413*	0.179*	0.178*	-0.098*	0.004
RBS14	0.533*	0.199*	-0.237*	-0.117*	0.125*
RBS15	0.061*	0.662*	0.016	0.011	0.099*
RBS16	0.053*	0.691*	0.125*	-0.005	0.03
RBS17	0.154*	0.511*	0.06	-0.013	0.021
RBS18	0.017	0.601*	0.135*	0.035	-0.042
RBS19	-0.024	0.437*	0.167*	0.090*	0.077*
RBS20	0.146*	0.337*	-0.162*	0.088*	0.279*
RBS21	0.003	0.438*	0.203*	0.219*	0.026
RBS22	0.181*	0.287*	0.311*	0.01	0.021
RBS23	0.023	0.240*	0.059*	0.274*	0.111*
RBS24	0.039	0.389*	-0.018	0.293*	0.162*
RBS25	0.016	0.452*	-0.062*	0.389*	0.038
RBS26	0.024	0.286*	0.04	0.467*	0.082*
RBS27	-0.002	0.235*	-0.031	0.372*	0.311*
RBS28	0	0.131*	-0.060*	0.279*	0.379*
RBS29	0.002	0.526*	-0.066*	0.372*	0.063*
RBS30	0.089*	-0.023	0.018	0.533*	0.102*
RBS31	0.135*	-0.100*	-0.01	0.590*	0.251*
RBS32	0.059	0.287*	0.242*	0.428*	-0.106*
RBS33	0.033	0.204*	-0.026	0.621*	-0.055*
RBS34	0.028	0.182*	0.027	0.556*	0.044
RBS35	-0.046	0.284*	0.219*	0.528*	-0.102*
RBS36	0.052*	0.122*	0.186*	0.276*	0.196*
RBS37	0.126*	-0.187*	-0.019	0.735*	0.203*
RBS38	0.016	0.006	0.042*	0.873*	-0.007
RBS39	-0.019	0.049*	-0.015	0.834*	-0.007
RBS40	0.027	-0.021	0.074*	0.119*	0.721*
RBS41	0.063*	0.100*	0.145*	0.116*	0.564*
RBS42	-0.026	0.135*	0.578*	-0.070*	0.382*
RBS43	-0.086*	0.125*	0.657*	-0.045*	0.257*

REFERENCES

Achenbach, T. & Rescorla, L. (2001). Manual for the ASEBA School-Age Forms & Profiles. Burlington, VT: University of Vermont, Research Center for Children, Youth, & Families.

Aman, M. & Singh, N. (1986). *Manual for the aberrant behavior checklist*. East Aurura, NY: Slosson Educational Publications.

American Psychiatric Association. (2000). Diagnostic and statistical manual of mental disorders (4th ed., text rev.). Washington, DC: Author.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Washington, DC: Author.

Barrett, S., Prior, M., & Manjiviona, J. (2004). Children on the borderlands of autism differential characteristics in social, imaginative, communicative and repetitive behavior domains. *Autism, 8,* 61-87.

Bauman, M. (1992). *Motor dysfunction in autism*. In: Joseph, A. & Young, R. (Eds.) Movement disorders in neurology and psychiatry. Boston: Blackwell; 660-663.

Bentler, P. (1990). Comparative fit indexes in structural models. *Psychological bulletin*, *107*(2), 238.

Bishop, S., Richler, J., & Lord, C. (2006). Association between restricted and repetitive behaviors and nonverbal IQ in children with autism spectrum disorders. *Child Neuropsychology, 4,* 247-267.

Bishop, S., Guthrie, W., Coffing, M., & Lord, C. (2011). Convergent validity of the mullen

scales of early learning and the differential ability scales in children with autism

spectrum disorders. *American Journal on Intellectual and Developmental Disabilities,*

116, 331-343.

Bishop, S., Hus, V., Duncan, A., Huerta, M., Gotham, K., Pickles, A., & Lord, C. (2013).

Subcategories of restricted and repetitive behaviors in children with autism

spectrum disorders. *Journal of autism and developmental disorders*, *43*(6), 1287- 1297.

Bodfish, J. (1995). Stereotypy, self-injury, and related abnormal repetitive behaviors.

Bodfish, J., Symons, F., Parker, D., & Lewis, M. (2000). Varieties of repetitive behavior in

autism: comparisons to mental retardation. *Journal of Autism and Developmental*

Disorders, 30, 237-243.

Bodfish, J. (2007). Stereotypy, self-injury, and related abnormal repetitive behaviors.

In *Handbook of intellectual and developmental disabilities* (pp. 481-505). Springer

US.

Bodfish, J., Symons, F., Parker, D., & Lewis, M. (2000). Varieties of repetitive behavior in

autism: Comparisons to mental retardation. *Journal of Autism and Developmental*

Disorders, 30, 237-243.

Bodfish, J. & Lewis, M. (2002). Self-injury and comorbid behavior in developmental,

neurological, psychiatric, and genetic disorders. In S. Schroeder, M. Oster-Granite, &

T. Thompson (Eds.). *Self-injurious behavior: Gene-brain-behavior relationships* (Vol.

2, pp. 23-39). Washington, DC: American Psychological Association Press.

Bodfish, J., Parker, D., Lewis, M., Sprague, R., & Newell, K. (2001). Stereotypy and motor

    control: differences in the postural stability dynamics of persons with stereotyped and

    dyskinetic movement disorders. *American Journal of Mental Retardation*, *106*(2):123–

    34.

Boyd, J., McBee, M., Holtzclaw, T., Baranek, G., & Bodfish, J. (2009). Relationships among

    repetitive behaviors, sensory features, and executive functions in high functioning

    autism. *Research in Autism Spectrum Disorders, 3*(4), 959-966.

Brown, J. (2009). Choosing the right type of rotation in PCA and EFA. *Shiken: JALT Testing

    and Evaluation SIG Newsletter, 13*(3), 20-25.

Browne, M. & Cudeck, R. (1993). Alternative ways of assessing model fit. *Sage focus

    editions*, *154*, 136-136.

Bryant, F. & Yarnold, P. (1995). Principal-components analysis and confirmatory factor

    analysis. In L.G. Grimm & P.R. Yarnold (Eds.), *Reading and understanding multivariate

    statistics* (pp. 99-136). Washington, DC: American Psychological Association.

Carpentieri, S. & Morgan, S. (1996). Adaptive and intellectual functioning in autistic and non-

    autistic retarded children. *Journal of Autism and Developmental Disorders, 28,* 611-620.

Cox, A., Klein, K. Charman, T., Baird, G., Baron-Cohen, S., Swettenham, J., et al. (1999).

    Autism spectrum disorders at 20 and 42 months of age: stability of clinical and adi-r

    diagnosis. *Journal of Child Psychology and Psychiatry, 40*(5), 719-732.

Cuccaro, M., Shao, Y., Grubber, J., Slifer, M., Wolpert, C., Donnelly, S., et al. (2003). Factor

    analysis of restricted and repetitive behaviors in autism using the autism diagnostic

    interview-r. *Child Psychiatry and Human Development, 34,* 3-17.

Damiano, C., Nahmias, A., Hogan-Brown, A., & Stone, W. (2013). What do repetitive and
stereotyped movements mean for infant siblings of children with autism spectrum
disorders? *Journal of Autism and Developmental Disorders, 43*(6), 1326-1335.

Dumont, R., Cruse, C., Price, L., & Whelley, P. (1996). The relationship between the
differential ability scales (DAS) and the wechsler intelligence scale for children- third
edition (WISC-III) for students with learning disabilities. *Psychology in the Schools, 33,*
203-209.

Elison, J., Wolff, J., Reznick, J., Botteron, K., Estes, A., Gu, H., ... & Piven, J. (2014). Repetitive
behavior in 12-month-olds later classified with autism spectrum disorder. *Journal of
the American Academy of Child & Adolescent Psychiatry*, *53*(11), 1216-1224.

Elliott, C. (2007). *Differential ability scales 2nd Ed*. New York: Harourt Assessment.

Esbensen, A., Seltzer, M., Lam, K., & Bodfish, J. (2009). Age-related differences in restricted
repetitive behaviors in autism spectrum disorders. *Journal of Autism and
Developmental Disorders, 39,* 57-66.

Evans, D., Leckman, J., Carter, A., Reznick, J., Henshaw, D., King, R., et al. (1997). Ritual,
habit and perfectionism: The prevalence and development of compulsive-like behavior in
normal young children. *Child Development, 68,* 58-68.

Evans, D., Lewis, M., & Iobst, E. (2004). The role of the orbitofrontal cortex in normally
developing compulsive-like behaviors and obsessive–compulsive disorder. *Brain and
cognition*, *55*(1), 220-234.

Fecteau, S., Mottron, L., Berthiaume, C., & Burack, J. (2003). Developmental changes of
autistic symptoms. *Autism, 6,* 67-81.

Finch, J., & West, S. (1997). The investigation of personality structure: Statistical models. *Journal of Research in Personality,* 31 (4), 439-485.

Frith, U. & Done, D. (1990). Stereotyped behavior in madness and in health. In S. Cooper & C. Dourish (Eds.), *Neurobiology of stereotyped behavior* (pp. 232-259). Oxford: Clarendon Press.

Gabriels, R., Cuccaro, M., Hill, D., Ivers, B., & Goldson, E. (2005). Repetitive behaviors in autism: relationships with associated clinical features. *Research in Developmental Disabilities, 26* (169-181).

Gadow, K., DeVincent, C., & Pomeroy, J. (2006). ADSD symptoms in children with pervasive developmental disorder. *Journal of Autism and Developmental Disabilities, 36*(2), 271-283.

Georgiades, S., Papageorgiou, V., & Anagnostou, E. (2010). Brief report: repetitive behaviors in Greek individuals with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 40,* 903-906.

Gidley-Larson, J. & Mostofsky, S. (2008). Evidence that the pattern of visuomotor sequence learning is altered in children with autism. *Autism Research, 1,* 341-353.

Gorsuch, R. (1983). Factor analysis (2nd ed.). Hillsdale, NJ: Lawrence Erlbaum Associates.

Gotham, K., Bishop, S., Hus, V., Huerta, M., Lund, S., Buja, A., & Lord, C. (2013). Exploring the relationship between anxiety and insistence on sameness in autism spectrum disorders. *Autism Research*, *6*(1), 33-41.

Haas, R., Townsend, J., Courchesne, E., Lincoln, A., Schreibman, L., & Yeung-Courchesne, R. (1996). Neurological abnormalities in infantile autism. *Journal of Child Neurology, 11,* 84-92.

Hallett, M., Lebiedowska, M., Thomas, S., Stanhope, S., Denckla, M., Rumsey, J. (1993). Locomotion of autistic adults. *Archives of Neurology, 50*(12); 1304-1308.

Harrop, C., McConachie, H., Emsley, R., Leadbitter, K., Green, J., & PACT Consortium. (2014). Restricted and repetitive behaviors in autism spectrum disorders and typical development: Cross-sectional and longitudinal comparisons. *Journal of autism and developmental disorders*, *44*(5), 1207-1219.

Harrop, C., Gulsrud, A., & Kasari, C. (2015). Does Gender Moderate Core Deficits in ASD? An Investigation into Restricted and Repetitive Behaviors in Girls and Boys with ASD. *Journal of autism and developmental disorders*, *45*(11), 3644-3655.

Hartigan, J. & Wong, M. (1979). Algorithm AS 136: A k-means clustering algorithm. *Journal of the Royal Statistical Society (Series C), 28*(1), 100-108.

Hattori, J., Ogino, T., Abiru, K., et al. (2006). Are pervasive developmental disorders and attention-deficit-hyperactivity disorder distinct disorders? *Brain and Development, 28*(6), 371-374.

Holtmann, M., Bolte, S., & Pourstka, F. (2007). Attention deficit hyperactivity disorder symptoms in pervasive developmental disorders: associations with autistic behavior domains and coexisting psychopathology. *Psychopathology, 40*(3): 172- 177.

Honey, E., McConachie, H., Randle, V., Shearer, H., & LeCouteur, A. (2008). One-year change in repetitive behaviors in young children with communication disorders including autism. *Journal of Autism and Developmental Disorders, 38*, 1439-1450.

Hu, L. & Bentler, P. (1999). Cutoff criteria for fit indexes in covariance structure analysis: Conventional criteria versus new alternatives. *Structural equation modeling: a multidisciplinary journal*, *6*(1), 1-55.

Inaba, M., Katoh, N., & Imai, H. (1994). Applications of weighted voronoi diagrams and

   randomization to variance-based k-clustering. *Proceedings of 10th ACM Symposium on*

   Computational Geometry; 332–339.

Joosten, A., Bundy, A., & Einfeld, S. (2009). Intrinsic and extrinsic motivation for stereotypic

   and repetitive behavior. *Journal of Autism and Developmental Disorders, 39*(3), 521-

   531.

Joseph, L., Thurm, A., Farmer, C., & Shumway, S. (2013). Repetitive behavior and restricted

   interests in young children with autism: comparisons with controls and stability over 2

   years. *Autism Research,* 6, 584-595.

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

Kessler, R., Brandenburg, N., Lane, M., et al. (2005). Rethinking the duration requirement

   for generalized anxiety disorder: evidence from the national comorbidity survey

   replication. *Psychological Medicine, 35*(7), 1073-1082.

Kim, S. & Lord, C. (2010). Restricted and repetitive behaviors in toddlers and preschoolers

   with autism spectrum disorders based on the autism diagnostic observation

   schedule (ADOS). *Autism Research, 3,* 162-173.

Kim, J. & Mueller, C. (1978). *Introduction to factor analysis: What it is and how to do it.*

   Beverly Hills, CA: Sage.

Kline, R. (2010). *Principles and practice of structural equation modeling (3rd ed.).* New York,

   New York: Guilford Press.

Kuhlthau, K., Orlich, F., Hall, T., Sikora, D., Kovacs, E., Delahaye, J., & Clemons, T. (2010).

   Health-related quality of life in children with autism spectrum disorders: results from

the autism treatment network. *Journal of Autism and Developmental Disorders,* *40,* 721-729.

Lam, K., & Aman, M. (2007). The repetitive behavior scale revised: independent validation

    in individuals with autism spectrum disorders. *Journal of Autism and Developmental*

    Disorders, 37, 855-866.

Lam, K., Bodfish, J., & Piven, J. (2008). Evidence for three subtypes of repetitive behavior in

    autism that differ in familiality and association with other symptoms. *Journal of Child*

    Psychology and Psychiatry, 49, 1193-1200.

Leekam, S., Prior, M., & Uljarevic, M. (2011). Restricted and repetitive behaviors in autism

    spectrum disorders: a review of research in the last decade. *Psychological Bulletin*, 1

    37(4), 562-593.

Lewis, M. & Bodfish, J. (1998). Reptitive behavior disorders in autism. Special issue:

    "Autism". *Mental Retardation Developmental Disabilities Research Review, 4,* 80-89.

Lewis, M., & Kim, S. (2009). The pathophysiology of restricted repetitive behavior. *Journal*

    of neurodevelopmental disorders, *1*(2), 114.

Lloyd, S. (1982). Least squares quantization in PCM. *IEEE Transactions on Information Theory;*

    28(2), 129-137.

Loftin, R., Odom, S., & Lantz, J. (2008). Social interaction and repetitive motor behaviors.

    Journal of Autism and Developmental Disorders, 38, 1124-1135.

Lord, C., Rutter, M., DiLavore, P., & Risi, S. (1999). *Autism diagnostic observation schedule*

    (ADOS). Los Angeles, California: Western Psychological Services.

Lord, C., Risi, S., Lambrecht, L, Cook, E., Leventhal, B., DiLavore, P., et al. (2000). The autism

    diagnostic observation schedule-generic: A standard measure of social and

    communication deficits associated with the spectrum of autism. *Journal of Autism*

    and Developmental Disorders, 30(3), 205-223.

Lord, C., Rutter, M., & Le Couteur, A. (2003). *Autism diagnostic interview-revised.* Los

Angeles, CA: Western Psychological Services.

Lovaas, I., Newsom, C., & Hickman, C. (1987). Self-stimulatory behavior and perceptual

reinforcement. *Journal of Applied Behavior Analysis, 20,* 45-68.

Marsh, H. W., Balla, J. R., & Hau, K. T. (1996). An evaluation of incremental fit indices: A

clarification of mathematical and empirical properties. *Advanced structural equation

modeling: Issues and techniques*, 315-353.

Matson, J., Kiely, S., & Bamburg, J. (1997). The effect of stereotypies on adaptive skills as

assessed with the DASH-II and Vineland Adaptive Behavior Scales. *Research in

Developmental Disabilities*, *18*(6), 471-476.

Matson, J., Rivet, T., Fodstad, J., Dempsey, T., & Boisjoli, J. (2009). Examination of adaptive

behavior differences in adults with autism spectrum disorders and intellectual

disability. *Research in Developmental Disabilities*, *30*(6), 1317-1325.

Matson, J., Rieske, R., & Williams, L. (2013). The relationship between autism spectrum

disorders and attention-deficit/hyperactivity disorder: an overview. *Research in

developmental disabilities*, *34*(9), 2475-2484.

Mayes, S., Calhoun, S., Murray, M., Ahuja, M., & Smith, L. (2011). Anxiety, depression, and

irritability in children with autism relative to other neuropsychiatric disorders and typical

development. *Research in Autism Spectrum Disorders*, *5*(1), 474-485.

Mazurek, M., Vasa, R., Kalb, L., Kanne, S., Rosenberg, D., Keefer, A., & Lowery, L. A. (2013).

Anxiety, sensory over-responsivity, and gastrointestinal problems in children with

autism spectrum disorders. *Journal of abnormal child psychology*, *41*(1), 165-176.

McDonald, R., Green, G., Mansfield, R., Geckeler, N., Gardenier, N., Anderson, J., Holcomb, W., & Sanchez, J. (2007). Stereotypy in young children with autism and typically developing children. *Research in Developmental Disabilities: A Multidisciplinary Journal, 28*(3), 266-277.

Militerini, R., Bravavaccio, C., Falco, C., Fico, C., & Palermo, M. (2002). Repetitive behaviors in autistic disorder. *European Child & Adolescent Psychiatry, 11*(5), 210-218.

Mirenda, P., Smith, I., Vaillancourt, T., Georgiades, S., Duku, E., Szatmari, P., et al. (2010). Validating the repetitive behavior scale-revised in young children with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 40*(12), 1521-1530.

Molloy, C., Dietrich, K., Bhattacharya, A. (2003). Postural stability in children with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 33*(6); 643-652.

Mooney, E., Gray, K., & Tonge, B. (2006). Early features of autism. *European child & adolescent psychiatry*, *15*(1), 12-18.

Moore, V. & Goodson, S. (2003). How well does early diagnosis of autism stand the test of time? Follow-up study of children assessed for autism at age 2 and development of an early diagnostic service. *Autism, 7*(1), 47-63.

Mosconi, M., Kay, M., D'Cruz, A., Seidenfeld, A., Guter, S., Stanford, L., & Sweeney, J. (2009). Impaired inhibitory control is associated with higher-order repetitive behaviors in autism spectrum disorders. *Psychological medicine*, *39*(09), 1559-1566.

Mullen, E. (1997). *Mullen scales of early learning*. Los Angeles: Western Psychological Services.

Muthen, B. & Muthen, L. (2001). Mplus User's Guide. Los Angeles, CA: Muthen & Muthen; 2001.

Muthén, B., DuToit, S., & Spisic, D. (2013). Robust inference using weighted least squares and quadratic estimating equations in latent variable modeling with categorical and continuous outcomes. 1997. *Unpublished technical report*.

Nuzzolo-Gomez, R., Leonard, M., Ortiz, E., Rivera, C., & Greer, D. (2002). Teaching children with autism to prefer books or toys over stereotypy or passivity. *Journal of Positive Behavior Interventions, 4,* 80-87.

Pedhazur, E. & Schmelkin, L. (1991). Measurement, design, and analysis: An integrated approach. *Hillsdale, NJ: Erlbaum*.

Piven, J., Harper, J., Palmer, P., & Arndt, S. (1996). Course of behavioral change in autism: A retrospective study of high-IQ adolescents and adults. *Journal of the American Academy of Child & Adolescent Psychiatry*, *35*(4), 523-529.

Prior, M. & MacMillan, M. (1973). Maintenance of sameness in children with kanner's syndrome. *Journal of Autism and Developmental Disorders, 3*(2), 154-167.

Rao, P., & Landa, R. (2014). Association between severity of behavioral phenotype and comorbid attention deficit hyperactivity disorder symptoms in children with autism spectrum disorders. *Autism*, *18*(3), 272-280.

Ravizza, S. & Ivry, R. (2001). Comparison of the basal ganglia and cerebellum in shifting attention. *Journal of Cognitive Neuroscience, 13,* 285-297.

Ravizza, S., Solomon, M., Ivry, R., & Carter, C. (2013). Restricted and repetitive behaviors in autism spectrum disorders: The relationship of attention and motor deficits. *Development and psychopathology*, *25*(03), 773-784.

Ray-Subramanian, C., & Weismer, S. (2012). Receptive and expressive language as

    predictors of restricted and repetitive behaviors in young children with autism

    spectrum disorders. *Journal of autism and developmental disorders*, *42*(10), 2113-2120.

Richler, J., Bishop, S., Kleinke, J., & Lord, C. (2007). Restricted and repetitive behaviors in

    young children with autism spectrum disorders. *Journal of Autism and*

    Developmental Disorders, 37(1), 73-85.

Richler, J., Huerta, M., Bishop, S., & Lord, C. (2010). Developmental trajectories of restricted

    and repetitive behaviors and interests in children with autism spectrum disorders.

    Development and Psychopathology, 22, 55-69.

Rojahn, J., Aman, M., Matson, J., & Mayville, E. (2003). The aberrant behavior checklist and

    behavior problems inventory: convergent and divergent validity. *Research in*

    Developmental Disabilities, 24, 391-404.

Rutter, M. (1996). Autism research: Prospects and priorities. *Journal of Autism and*

    Developmental Disorders, *26*(2), 257-275.

Rutter, M., Le Couteur, A., & Lord, C. (2003). *Autism diagnostic interview-revised (ADI-R)*. Los

    Angeles, California: Western Psychological Services.

Ruzzano, L., Borsboom, D., & Geurts, H. (2014). Repetitive behaviors in autism and

    obsessive-compulsive disorder: new perspectives from a network analysis. *Journal*

    of Autism and Developmental Disorders.

Satorra, A., & Bentler, P. (2001). A scaled difference chi-square test statistic for moment

    structure analysis. *Psychometrika*, *66*(4), 507-514.

Schooler, J. (2014). Metascience could rescue the replication crisis'. *Nature*, *515*, 9.

Shafran, R. (1998). Childhood obsessive-compulsive disorder. In P.J. Graham, (Ed),
    Cognitive-behavioral therapy for children and families. New York: Cambridge
    University Press.

Sparrow, S., Cicchetti, D., & Balla, D. (2005). *The vineland II adaptive behavior scales*. Circle
    Pines, MN: American Guidance Service.

Sugar, C. A., & James, G. M. (2011). Finding the number of clusters in a dataset. *Journal of the
    American Statistical Association*.

Suhr, D. (2006). Exploratory or confirmatory factor analysis? *Statistics and Data Analysis*, *31*.

Szatmari, P., Georgiades, S., Bryson, S., Zwaigenbaum, L., Roberts, W., Mahoney, W., et al.
    (2006). Investigating the structure of the restricted, repetitive behaviors and
    interests domain of autism. *Journal of Child Psychology and Psychiatry, 47*(6), 582-
    590.

Thelen, E. (1979). Rhythmical stereotypies in normal human infants. *Animal Behavior, 27,*
    699-715.

Thelen, E. (1981). Rhythmical behavior in infancy: an ethological perspective.
    Developmental Psychology, 17, 237-257.

Thompson, B. (2004). Exploratory and confirmatory factor analysis: understanding concepts and
    applications. Washington, DC, US: American Psychological Association.

Tonge, B., Brereton, A., Gray, K., & Einfeld, S. (1999). Behavioral and emotional disturbance
    in high-functioning autism and Asperger syndrome. *Autism, 3,* 117-130.

Troyb, E. (2014). Restricted and repetitive behaviors as predictors of outcome in autism
    spectrum disorders. *Doctoral Dissertations,* 520.
    http://digitalcommons.uconn.edu/dissertations/520

Turner, M. (1995). *Repetitive behavior and cognitive functioning in autism.* Unpublished

    doctoral thesis, University of Cambridge, UK.

Turner, M. (1997). Towards and executive dysfunction account of repetitive behavior in autism.

    In J. Russel (Ed.), *Autism as an executive disorder* (pp. 57-100). Oxford: Oxford

    University Press.

Turner, M. (1999). Annotation: Repetitive behavior in autism: A review of psychological

    research. *Journal of Child Psychology and Psychiatry, and Allied Disciplines, 40*(6),

    839-349.

Volkmar, F., Sparrow, S., Goudreau, D., & Cicchetti, D. (1987). Social deficits in autism: an

    operational approach using the vineland adaptive behavior scales. *Journal of the*

    American Academy of Child and Adolescent Psychiatry, 26, 156-161.

Watt, N., Wetherby, A., Barber, A., & Morgan, L. (2008). Repetitive and stereotyped

    behaviors in children with autism spectrum disorders in the second year of life.

    Journal of Autism and Developmental Disorders, 38(8), 1518-1533.

Wechsler, D. (1999). *Wechsler abbreviated scale of intelligence.* San Antonio, TX:

    Psychological Corporation.

Weschsler, D. (2003). *Wechsler intelligence scale for children: fourth edition.* San Antonio,

    TX: Psychological Corporation

Wilens, T., Biederman, J., & Spencer, T. (2002). Attention deficit/hyperactivity disorder across

    the lifespan. *Annual Review of Medicine, 53*, 113-131.

Wolff, J., Botteron, K., Dager, S., Elison, J., Estes, A., Gu, H., ... & Zwaigenbaum, L. (2014).

    Longitudinal patterns of repetitive behavior in toddlers with autism. *Journal of Child*

    Psychology and Psychiatry, *55*(8), 945-953.

Xu, S., Qiao, X., Zhu, L., Zhang, Y., Xue, C., & Li, L. (2016). Reviews on Determining the Number of Clusters. *Applied Mathematics and Information Sciences*, *10*(4), 1493-1512.

Yerys, B., Wallace, G., Sokoloff, D., et al. (2009). Attention deficit/hyperactivity disorder moderate cognition and behavior in children with autism spectrum disorders. *Autism Research, 2*(6), 322-333.

Yu, C.(2002). Doctoral dissertation. Los Angeles: University of California; 2002. Evaluating cutoff criteria of model fit indices for latent variable models with binary and continuous outcomes.

Zandt, F., Prior, M., & Kyrios, M. (2007). Repetitive behavior in children with high functioning autism and obsessive compulsive disorder. *Journal of Autism and Developmental Disorders, 37,* 251-259.