

EXPLORATION OF GENDER INFLUENCES IN RESTRICTED AND REPETITIVE
BEHAVIORS IN CHILDREN WITH AUTISM SPECTRUM DISORDER

by

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ABSTRACT

More boys than girls have ASD at a ratio of 4:1 (Baio et al., 2018). In general, boys tend to demonstrate more ritualistic and repetitive behaviors (RRBs) than girls (Szatmari et al., 2012) though a gender bias may lead to under diagnosis of girls. Therefore, a female phenotype should be differentiated to improve diagnostic accuracy.

This study seeks to describe and compare RRB profiles and frequencies in two age cohorts of girls and boys with ASD as measured by the Repetitive Behavior Scale-Revised (RBS-R) and the Childhood Routines Inventory (CRI).

214 children with ASD (42 girls; 172 boys) between 16 months and 10 years old (mean = 46.1 months) were included from an ASD clinical research database. Two age cohorts were used (1-3 years; 4-10 years) to compare RRBs using parent reports.

Girls younger than 3 had more repetitive ($t = -1.66$; $p = .03$), ritualistic ($t = -.12$; $p = .004$), and sameness behaviors ($t = -1.9$; $p = .002$). Older girls had more stereotyped behaviors than boys ($t = -2.2$; $p = .001$) and more ritualistic behaviors in general. Younger girls ($n = 23$) had more stereotyped (62.7%), restricted interests (58%), sameness (41%), compulsive (46%), ritualistic (36%), and self-injurious behavior (27%). Boys displayed RRBs and stereotypical behaviors. Girls demonstrate more repetitive behaviors than boys. RRB patterns and profiles will be discussed comparatively. These findings contribute the understanding of the female ASD phenotype.

DEDICATION

This thesis is dedicated to my family who supported me through my first writing and research experience. I would like to thank my grandmother, Catherine Arias, whose sacrifice and dedication to education and family paved the way for my own successes. From her I learned the importance of tenacity, faith, and a strong work ethic. I can because she did. I could not have completed this process without the steadfast support of my dad. Two thousand miles is not all that far when home is a person. Because of him I know that acceptance is the answer to all of my problems today. Finally, I dedicate this paper a mi mamá quien es la mejor ejemplo de la fortaleza. Somos como dos gotas de agua.

LIST OF ABBREVIATIONS AND SYMBOLS

<i>ASD</i>	Autism Spectrum Disorder
<i>DD</i>	Developmental delay
<i>RRB</i>	Restricted and repetitive behavior
<i>TD</i>	Typical development
<i>t</i>	Computed value of <i>t</i> test
=	Equal to

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CHAPTER 1 INTRODUCTION

Autism Spectrum Disorder (ASD) is a neuro-developmental disability characterized by difficulties in communication and interaction and the presence of repetitive and ritualistic behavior (American Psychiatric Association [APA], 2013). Early studies on children with ASD document minimal interactions with other people and a strong preference toward objects (Kanner, 1943). ASD is one of the most extensively researched disorders, partially due to the large population that fall within the spectrum it describes. In 2000 approximately 1 in 150 children were identified with ASD and in 2012 the number dramatically increased to 1 in 68 children receiving a diagnosis (Christensen et al., 2016). Recently, the Center for Disease Control and Prevention (CDC) released the most updated figures with 1 in every 59 children being identified with ASD (Baio et al., 2018). One explanation for the increased prevalence points to changes in description and characteristics that fit under the umbrella term ASD. The fifth and most current edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-V) was released in 2013 and redefined characteristics of ASD with two cornerstone traits, deficits in social communication and social interaction and the presence of restricted repetitive patterns of behavior (APA, 2013). The easiest for clinicians and medical persons to observe are restricted and repetitive behaviors (RRBs). These mannerisms are distinct and reasonably easy to evaluate (Turner, 1999). Though RRBs are the most salient behaviors, they are not necessarily the most reliable during the evaluation. For example, one type of RRB, restricted behavior, is used to describe individuals with a limited range of interests. A child may demonstrate a restricted interest, however, if the object is not present during the evaluation process, this behavior will not be taken into diagnostic consideration. Therefore, parent reports of RRB may

be valuable to the diagnostic process. Early intervention resources and diagnostic materials are now more effective than ever thanks to increased knowledge and early symptom presentation in ASD. Parent report and the interview process are extremely valuable tools when documenting behaviors that may not manifest in a testing environment. Extensive research has explored social communication in ASD, though there is still relatively little information known about RRBs (Harrop et al. 2014), including the variability and influence of gender.

Social Communication

Social communication refers to the ability to use language in collective or group situations. Components of social communication incorporate social cognition (Theory of Mind), social interaction (speech styles), pragmatics (verbal and nonverbal communication), and language processing (spoken and written language management) (Grice, 1975; Nelson, 1978; Timler, G. R., Olswang, L. B., & Coggins, T. E., 2005).

These abilities allow pragmatic skills to be effective and successful during daily interactions. Effective social communicators can coordinate eye gaze, gestures, words, balance of turns and demonstrate motor imitation ability to communicate with others (Bates, Benigni, Bretherton, Camioni, & Volterra, 1979; Wetherby & Prutting, 1984)

Social Communication in Typical Development

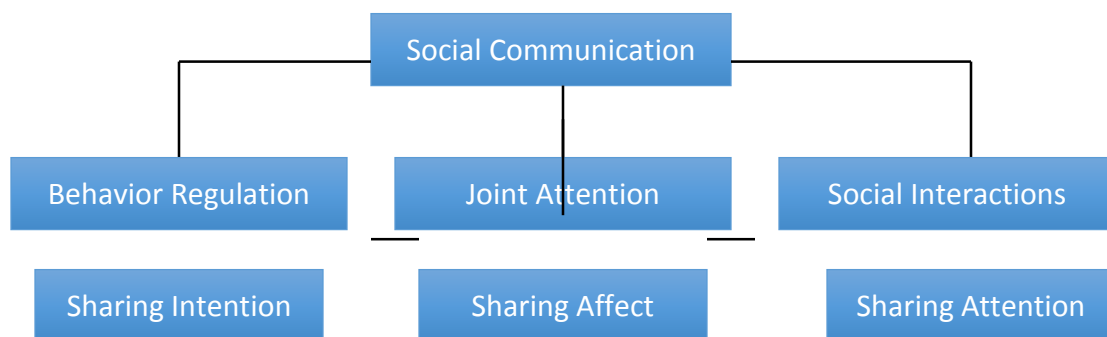
Communicative intention is a term used to describe purposeful interaction. Communication is goal directed and an individual's level for interaction can be separated into three categories: perlocutionary, illocutionary, and locutionary stages. Between birth to eight months of age, typically developing children fall within the perlocutionary stage. This type of interaction consists of crying, cooing, and utilizes nonspecific use of body and face. At this point, acts are treated as if they are communicative (Paul, 2010). During the perlocutionary stage

infants sustain interactions or respond to stimuli. This second stage of communication intention develops between eight to twelve months of age. It is characterized by intentional communication including: gesturing, gaze, and vocalizations. Locutionary communication is the last stage of development that takes place between 12-18 months of age. At this time first words are spoken and the same communicative intent expressed via gestures utilizes words instead (Bates, Camaoini, & Volterra, 1975; Paul, 2010; Wetherby & Prizant, 1989).

Non-verbal communicative functions are described in three categories: behavior regulation, joint attention, and social communication. Though these functions are first used through non-word vocal productions, they are later used through language following word development (Bruner 1981; Wetherby & Prizant 1993). Behavior regulation refers to requests of objects and actions, or protesting (e.g. reaching for a toy, pushing a hand away to protest it). Social interactions are used for attention seeking purposes. They consist of representation gestures, social games, and routine like interactions (e.g. playing patty-cake, waving “hello”). Joint attention is a communication category that relies on social protocols to direct other’s attention with the purpose of sharing an interest in an object or event (showing or point to a toy). It is believed that these communicative functions are related to and have an influence on social cognitive skills including the ability to distinguish between self and other people as well the understanding the use of others as a mean of attainment (Werner & Kaplan, 1963).

Across the first year of life a propensity for joint attention is delineated by social accomplishments and successfully shared experiences. See Figure 1. These skills include: sharing attention, sharing affect, and sharing intentions (Stern, 1985).

Figure 1. Social communication in early development.



The contribution these abilities make to social interaction is important to mention in typically developing (TD) children because they provide insight into the aspects of social communication that are most challenging for children with ASD. In fact researchers argue that functions, such as joint attention, are the basis of development for language and more intricate abilities like pretend play and theory of mind (Bruner, 1983; Charman, 1997; 2003; Carpenter, Nagell, Tomasello, Butterworth, & Moore, 1998).

Social Communication in ASD

As mentioned previously, deficits in social communication skills are a hallmark symptom of ASD. Indications of ASD, pertaining to social communication skills, present in young children in a variety of ways. Age, gender, cognitive abilities, and comorbid diagnoses are factors that influence the type and severity of social communication disorders (Lainhart, 1999; Fombonne 2003). Impacted skills include: decreased eye gaze, lack of joint or shared attention, decreased social or reciprocal smiling, low use of gestures, limited assimilation of eye gaze, facial expression, gestures, and vocalization with intention of communication (Zwaigenbaum et al., 2009). Domains of social communication impairments in ASD have been established and supported in research. Three elements consistently identified in this demographic are: joint

attention, affective reciprocity, and theory of mind (Mundy, Sigman, & Kasari 1990; Lord, 1997; Robertson, Tanguay, L'ecuyer, Sims, & Waltrip, 1999; Tanguay, Robertson, & Derrick, 1998).

Impairments in joint attention are commonly associated in children with ASD. Whereas their TD peers communicative functions are represented in the three categories mentioned above (behavior regulation, joint attention, and social communication), children with ASD communicate primarily for behavioral regulation purposes (Wetherby & Prutting, 1984). A number of researchers studying social communication in this population found that impairments in proto-declarative (sharing or showing behaviors) attention are more severely affected than proto-imperative attention (requesting behaviors) (Charman, 1998; Mundy, Sigman, Kasari, 1990; Mundy, Sigman, Ungerer, & Sherman 1986; 1987). Children with ASD also exhibit deficits of symbol use. These limitations include use of language, gestures, play, and imitation.

Social Communication Differences in ASD vs TD

Though challenging to distinguish, communicative intentions between ASD and TD profiles could not be more different. Researchers suggest that before 12 months children with TD communicate before the presence of words for the purposes of behavior regulation, social interaction, and joint attention. (Bruner, 1981; Wetherby & Prizant 1993). On the other hand, children with ASD communicate for behavior regulation purposes and struggle with joint attention (Stone & Caro-Martinez, 1990; Wetherby, Prizant, & Hutchinson, 1998; Wetherby, Watt, Morgan, & Shumway, 2007). Kanner (1943) explained these differences best in his research exploring early cases of ASD. He wrote that the deficit in these abilities was found in “the children’s inability to relate themselves in the ordinary way to people and situations from the beginning of life” (p. 242).

The acquisition of spoken language is an exceptionally accurate predictive measure of positive outcomes in ASD (Toth, Munson, & Meltzoff, 2006). An interesting difference between children with ASD and TD are the rates at which social communication skills progress with respect to severity and age of diagnosis. Social communication development was described and compared between children identified with ASD early in life, children with a later ASD diagnoses, and non-ASD peers. Results of the study found that at 14 months, social communication abilities between the group with the later diagnoses and non-ASD peers were almost identical. However, by 24 months all ASD groups had regressed in social behavior compared to other groups. Impairments in shared positive affect and reduced inventory of gestures were specifically noted (Landa, Holman, & Garrett-Mayer 2007).

Twenty-four months appears to be the point at which ASD profiles become noticeably different from their age related peers within communicative measures. Wetherby et al., (2007) explored social communication between children with ASD, TD, and developmental delay (DD). Five deficits unique to ASD were cited in this study. These characteristics appeared between age 18-24 months and include: gaze shifts, gaze/point follow, rate of communicating, acts for joint attention, and inventory of gestures (Wetherby et al., 2007 p. 973).

Repetitive Behaviors

RRBs are a hallmark of ASD diagnosis. These behaviors were some of the first noted traits in primary accounts of the disorder (Kanner, 1943). The children studied in Kanner's early research present with behavior that is "anxiously obsessive desire for the maintenance of sameness that nobody but the child may disrupt on rare occasions" (p. 245). This insistence on sameness described by Turner (1999) describes these behaviors in two ways using a paradigm often cited in current ASD literature. Lower-level behaviors are categorized by repetitive

movements, they consist of stereotyped mannerisms, tics, and self-injurious repetitive behaviors. Studies have found these lower-level behaviors most frequently in children with developmental delays, issues with pragmatic language, and intellectual disabilities (Barrett, Prior, & Manjiviona, 2004; Lam, Bodfish, & Piven, 2008).

Though a cornerstone of a diagnosis, RRBs are not exclusive to ASD. In fact, RRBs are found in a variety of children including those with TD and as well as intellectual disabilities and language disorders (Evans 1997; Sallustro & Atwell 1978; Thelen, 1979). A number of studies discuss the implications of RRBs on learning and developmental outcomes. RRBs have been proven to negatively impact the learning and socialization process of individuals with ASD (Koegel & Covert, 1972; Loftin, Odom, & Lantz, 2008).

However other researchers discuss RRBs in terms of coping strategies used during anxiety inducing social situations (Baron-Cohen, 1989). This claim suggests that RRBs may be purposeful for some children though not consistently so for all. Other literature explores repetitive behaviors as predictors for later developmental outcomes. In a study examining RRBs in children with ASD, Watt, Wetherby, Barber, & Morgan (2008) found that RRBs with objects were related to measures of social competence and acted as predictors of developmental outcomes within the second year of life. Additionally research has found that disruption and negative impacts in family functioning are consequences of RRBs. RRBs interfere with learning and functioning (Koegel & Covert, 1972; Pierce & Courchesne, 2001) are identified as stressful behaviors for caregivers (Bishop, Richler, Cain, & Lord, 2007).

Higher-level behaviors include insistence on sameness, attachment to objects, and confined interests (Turner, 1999). New research suggests that due to a genetic foundation, these

behaviors may run in families more often than lower order behaviors (Boyd, McDonough, & Bodfish, 2012).

Repetitive behaviors are not strictly limited to individuals with ASD. In fact these qualities are found in children with typical development (TD) (DeLoache, Simcock, & Macari, 2007) and developmentally delayed (DD) children as well. Watt et al., (2008) conducted a study comparing RRBs in groups of ASD, TD, and DD children between 18-24 months. They found that the ASD group displayed repetitive behaviors more often and for longer periods of time than both TD and DD children.

Nonverbal cognitive skills (NVIQ) often compromise aspects of social communication, outlined in the diagram above, combined with visuospatial, and fine motor abilities. Among researchers, however, there appears to be a disconnect when distinguishing the relationship between NVIQ and RRB prevalence. Recent studies present a compelling argument that describe social communication and RRBs as separate and unrelated domains of ASD, two processes that processes develop independently.

Conversely others studies have found an inverse relationship between the two domains (Gabriels, Cuccaro, Hill, Ivers, & Goldson, 2005; Paul, Chicchetti, & Volkmar, 2008; Ray-Subramanian & Ellis Weisner, 2012). Ray-Subramanian (2012) examined the relationship between language skills and nonverbal cognitive skills (NVIQ), and RRBs for children on the spectrum between ages 2 to 3 years old. Results supported a negative correlation between receptive skills and RRBs. There are contradictory findings between researchers regarding the congruency between RRBs and social communication abilities. Boyd et al., (2012) summarizes this conflicting relationship in a concise manner:

Overall findings on the relationship between IQ and repetitive behaviors appear to indicate that cognitive and social-communication deficits do not fully account for the expression of repetitive behaviors in ASD...(p. 3)

It is important to discuss the relationship between NVIQ and repetitive behaviors. NVIQ is indicative of, at a minimum, some aspects of social communication and communicative abilities. There is evidence that social communication abilities are improved when RRBs, behaviors related to IQ, have decreased. NVIQ and RRBs feed into and provide insight toward social communication abilities.

Gender Differences in ASD

According to the CDC, ASD occurs significantly more often in boys compared to girls, with a ratio of 4:1 (Baio et al., 2018) respectively. One explanation for this ratio is that girls are simply being missed during the diagnostic evaluation process. Dworzynski, Ronald, Bolton, & Happé (2012) compared children aged 10-12 with an ASD diagnosis to others who failed to receive a diagnosis regardless of their severity score on the *Childhood Autism Spectrum Test* (CAST). Researchers found that girls were less likely than boys to receive an ASD diagnosis despite similar scores on the CAST; however girls who did meet diagnostic criteria displayed comorbid concerns such as low intellectual levels and behavioral difficulties. This suggests an unintentional gender bias in ASD testing as well potentially superior coping skills in females with ASD.

The gender ratio has resulted in researchers attempts to identify and describe how ASD presents between males and females. So far investigators have agreed on several key differences. First, there are more males than females diagnosed with ASD. Second, intellectual ability appears to be a trend between genders as well. Females with ASD tend to have higher IQ scores

and better social communication abilities than boys (Halladay et al., 2015). Finally, females appear to engage in fewer repetitive behaviors compared to boys (Szatmari et al., 2012). However, many agree that the quality, not the quantity, of repetitive behaviors is where females differ. Halladay et. al (2015) presents a salient example of this difference from the perspective of an individual with ASD:

For example, a young woman with autism noted that carrying several well-worn books everywhere she goes, and constantly reading them to the detriment of all other social interactions, may be repetitive behavior that goes undetected. Clinicians noted that circumscribed interests around dolls or babies in females might be misinterpreted as pretend play... (pg 2)

As mentioned previously, intellectual ability may also contribute to the discrepancy when examining the gender ratio in ASD. Research reports disparities when answering the question of whether females display fewer socio-communication difficulties compared to males. Some studies have not found gender differences in social behavior (Banach et al., 2009; Carter, Black, Tewani, Connolly, Kadlec, & Tager-Flusberg, 2007; Van Winjngaarden-Cremers et al., 2014). Conversely, several researchers report that social play and imitative play do indicate a gender bias (Tsai & Beisler 1983; Holtmann, Bölte, & Poustka, 2007). The inconsistencies in this line of research is indicative of the need for additional testing in IQ, developmental abilities, and comorbidities to accurately characterize females with ASD (Volkmar, Szatmari, & Sparrow, 1993; Harrop et al., 2015. Van Winjngaarden-Cremers et al., (2014) explains that the current ASD definition is created in the image of the male phenotype. The researchers recommended a reevaluation of female phenotypes of ASD in hopes of creating a diagnostic tool that is more sensitive to a female profile.

It is important to review and assess the reliability of using RRBs to diagnose ASD in females, which may present differently than they do in males. For example, extremely intense interests (EII), a characteristic of repetitive behaviors in ASD, have been found to be more prevalent in boys than they are in girls (DeLoache, Simcock, & Macari, 2007).

Purpose

The primary purpose of this study is to explore the differences in repetitive behaviors with regard to sex differences in children with ASD. The following objectives will be addressed:

1. *Describe repetitive behavior profiles in girls compared to boys in children with autism.*
 - a. *Hypothesis 1: Based on previous research and clinical observation, a higher frequency and repertoire of repetitive behaviors in boys than girls is anticipated. More instances of lower order repetitive behaviors in females who have lower cognitive abilities compared to their male counterparts of equal or greater cognitive abilities are expected as measured by standardized tests including the Mullen Scales of Early Learning (MSEL), Wechsler Intelligence Scale for Children (WISC), and Differential Abilities Scale (DAS) administered according to age.*
2. *Examine the differences found in repetitive behaviors between genders.*
 - a. *Repetitive behaviors will be observed in all groups in the birth to five ranges; however we expect females to engage in more lower-level repetitive behaviors.*
 - b. *Based on previous research and clinical observation it is expected that there will be fewer repetitive behaviors in the older group, though these characteristics will still be present, specifically females in general with ASD. We expect RRBs and*

social communication abilities to develop at parallel rates, but overall have no influence on each other

CHAPTER 2 METHODS AND PROCEDURE

Participants

Participants included One hundred and seventy-five children (143 boys, 32 girls) who were recruited from The University of Alabama Autism Spectrum Disorders Clinic Database. The University of Alabama Autism Spectrum Disorders Clinic provides a comprehensive evaluation by a multidisciplinary team including a speech language pathologist, clinical psychologist, and a pediatrician. At the time of evaluation, parents are invited to participate in an ongoing research project titled: Autism Spectrum Disorders Clinic Research Database (IRB #06-OR-022-R7). The primary investigator and supervising faculty member are included as project personnel.

This research database was designed to allow for longitudinal study of autism symptomology, presentation, and change over time in a cohort of the sample. The database contains up to 250 variables per child. If the family consents to participate, their evaluation data is entered into the secure database, which may be accessed by research personnel and approved collaborators to answer specific research questions. Data are de-identified to protect participant confidentiality.

To be included in this study the following criteria were met by each participant: (1) a clinical diagnosis of ASD (2) between months of age 2 and 10 years at the time of their evaluation and (3) no co-morbid medical diagnosis that could attribute to repetitive behavior.

Procedures

The UA ASD Clinic has granted permission to allow access to the clinical research database to address these proposed questions. The primary researcher was added to the research database IRB. Participant data for children who met inclusion criteria were selected for the current study.

Measures

Repetitive Behavior Scale Revised (Bodfish, Symon, & Lewis, 1999).

The *Repetitive Behavior Scale-Revised* (RBS-R) is a parent reported questionnaire indicating the severity of RRBs in ASD. It consists of 43 items from 6 domains titled: stereotyped, self-injurious, compulsive, ritualistic, sameness, and restricted which are rated on a four-point Likert scale ranging from ‘behavior does not occur’ to ‘behavior occurs and is a severe problem’.

Measures include items endorsed and frequency, which yield two scores for repetitive behaviors; overall number of items endorsed and overall scores.

Childhood Routines Inventory.

The *Childhood Routines Inventory* (CRI) is a parent reported questionnaire that measures 19 RRB and compulsive-like behaviors along a 5-point Likert scale. Responses contribute to “just right” and “repetitive behaviors” composites. The questionnaire takes approximately 5 minutes to complete and yields a score for each of the 19 items, as well as several composite scores.

Autism Spectrum Rating Scale.

The *Autism Spectrum Rating Scale* (ASRS) is ASD rating scale given to caregivers and teachers. Using a five-point Likert rating scale, the ASRS is used to identify symptoms, behaviors, and associated features of ASD between ages 2 -18.

Autism Diagnostic and Observation Schedule (Lord, Rutter, DiLavore, & Risi, 2008).

The *Autism Diagnostic and Observation Schedule* (ADOS) is a semi-structured, play-based assessment with used to diagnose and assess social communication, play, and repetitive behavior. The ADOS presents a series of communication temptations and social presses related to core diagnostic criteria and scored relative to associated behaviors. These scores contribute to calibrated domain scores for two dimensions: Social Affect (SA) and Repetitive and Restricted Behavior (RRB). The SA domain score was used as the social measure for correlation analyses. These scores are appropriate for comparison across and within research samples. The algorithm used for the SA score provides an estimate of ASD-related social communication deficits as judged by a trained clinician (Hus, Gotham, & Lord, 2012).

Childhood Autism Rating Scale- Second Edition (Schopler, Bourgondien, Wellmand, & Love, 2010).

The *Childhood Autism Rating Scale* (CARS-2) is a behavioral rating scale used for assessing the presence and severity of symptoms of ASD. The assessment is used to observe and rate 15 items to yield a composite score indicating ASD severity.

The Mullen Scales of Early Learning (Mullen, 1995).

The *Mullen Scales of Early Learning* is a standardized developmental test for children between ages 3 to 69 months. It is used to measure early cognitive and motor development, visual reception, and expressive/receptive language.

Wechsler Intelligence Scale for Children.

The *Wechsler Intelligence Scale for Children* (WISC) is an individually administered intelligence test for children between the ages of 6 and 16. Upon completion the WISC generates

a Full Scale IQ that represents a child's general intellectual ability as well as the representative ability of discrete cognitive domains.

Differential Ability Scales (Elliott, Murray, & Pearson, 1990).

The *Differential Ability Scales* is an individually administered battery of cognitive and achievement tests intended for children ages 2-17.

CHAPTER 3 RESULTS

One hundred and seventy-five children (143 boys, 32 girls) were included in the final analysis. Ages ranged from 16 months to 124 months with a mean age of 42 months (boys= 43.5; girls= 40.21). The first aim of this research study focuses on describing RRB profiles by gender utilizing the RBS-R and the CRI.

RRBs measured by the RBS-R are composed of 6 behavior subdomains. These behaviors include: stereotyped, self-injurious, compulsive, ritualistic, sameness, and restricted interests.

The RBS-R is measured in terms of endorsed and total scores. Based on the RBS-R, girls demonstrated a higher overall frequency of RRB than boys.

Regarding frequency scores girls demonstrated more stereotyped behaviors than boys (girls mean= 9.17; boys = 6.24). Girls demonstrated more self-injurious behaviors (girls mean= 4.68; boys mean= 4.18); compulsive behaviors (girls mean= 7.06; boys mean= 6.31); ritualistic behaviors (girls mean= 5.10; boys mean=.03); and sameness behaviors than boys (girls mean= 9.97; boys mean= 8.32). Boys demonstrated more restricted interests (girls mean= 4.71; boys mean= 5.01). Overall, girls demonstrated a higher RBS-R overall total than boys (girls mean= 41.65; boys mean= 34.96). The group of girls revealed a greater overall mean as well as a greater standard deviation than the boys (girls SD= 31.34; boys SD= 26.99).

The pattern identified for the RBS-R totals was mirrored by the RBS-R items endorsed with two exceptions. More items were endorsed by boys for self-injurious behaviors (girls mean = 2.26; boys = 2.49) and ritualistic behaviors (girls = 2.52; boys = 2.63). That is, girls

demonstrated a higher frequency of self-injurious and ritualistic behavior, although boys demonstrated a larger repertoire of both of these types, though only slightly so.

This variation is reflective of the heterogeneous nature of ASD. The high SD and relative broad range in RBS-R overall total scores indicates that RRB profiles are variable among children with ASD. A summary of the means, standard deviations, and differences for RBS-R endorsed and total items are shown below in Table 1. Visual analyses are provided to illustrate differences across the RBS-R subdomains for endorsed and total items (see Figures 2 and 3).

The CRI was used as a measure of RRBs also based on parent report. CRI response composites include “just right” and “repetitive behavior” descriptors. Girls demonstrated a slightly larger number of CRI- just right behavior than boys (girls mean= 12.67; boys mean= 12.65). Additionally, girls revealed a higher number of repetitive behaviors than boys on the CRI- repetitive behavior index (girls mean= 11.27; boys mean= 11.05). A summary of the means, standard deviations and Visual analyses are provided to illustrate differences across CRI domains (see Table 1). Visual analysis is provided for CRI items (see Figure 4).

Statistical analysis was completed using R (R Core Team, 2012) to perform a linear fixed effects analysis to explore relationships and predictive value of items. As fixed effects we included: age, gender, ADOS SA total, CRIt, and global functioning. Analysis indicated that there was a relationship between CRIt and RBS-R overall totals as well as endorsed totals. However, it was noted that only the CRIt and RBS-R overall total were correlated and indicated a predictive function. Correlation coefficients and fixed effects can be found in Tables 4 and 5.

A central aim of this study was to identify whether an RRB profile emerged based on parent report, which could hold important diagnostic implications. Therefore, follow up analyses were conducted with children 36 months and younger, the age during which early diagnosis is

most likely. This sub sample consisted of 19 girls (mean age = 29 months) and 70 boys (mean age = 28.42). The RRB pattern was similar as in the full sample with the exception of sameness, a behavior that a greater number of girls demonstrated. Sameness, a higher order behavior according to Turner (1999), refers to an aversion to change and insistence of routine and order. Six percent more girls in the toddler sub sample (95%) engaged in this higher order behavior compared to boys (89%). This trend mirrors the means of the boys and girls in the sample that included all children who participated in the study and is reflected in the sameness subscale (see Table 1).

In the younger cohort, developmental ability as measured by the Mullen was inversely correlated with the RBS-R Endorsed ($r = -.313$; $p = .009$) RBS-R Total ($r = -.271$; $p = .024$) in boys but not in girls (Endorsed: $r = .194$; $p = .43$; Total: $r = .020$; $p = .94$).

Given the large range of behaviors demonstrated in both the full and reduced samples, percentages of behaviors endorsed by boys and girls were calculated to provide an alternate way to compare gender profiles. This analysis revealed an interesting pattern (see Table 2). Fewer girls demonstrated lower order behaviors than boys. Specifically, fewer girls demonstrated self-injurious, stereotyped, and restricted behaviors than boys. Fewer girls also demonstrated ritualistic behaviors, which is a higher order behavior, though marginally so.

CHAPTER 4 DISCUSSION

The first purpose of this paper was to describe RRB in a sample of children with ASD (32 girls, 143 boys). The RBS-R and CRI were used to measure instances and the severity of RRBs in children with ASD. It is important to note that repetitive behavior can be difficult to measure by therapists who are unfamiliar with the child. It is especially challenging when RRBs include objects that are not present in the evaluation environment and are only exhibited in the home. The presence of RRB in at least two diagnostic categories including stereotyped or repetitive movements, insistence on sameness, fixated or intense interests, or hyper/hypo response to sensory information is required for a diagnosis of ASD (APA, 2013). The DSM-V criteria specifies that the RRB can be observed or reported, emphasizing the need to better understand how parent report can be incorporated into the diagnostic process. In this study, caregiver report allowed for a comprehensive view into the interests and behaviors of their children. Regarding RBS-R total scores, girls demonstrated more stereotyped behaviors, self-injurious behavior, compulsive behavior, ritualistic behavior, and sameness behavior than boys. Though boys demonstrated a greater frequency of restricted interests, girls had a higher RBS-R overall total. Regarding RBS-R endorsed items, girls demonstrated higher scores in all categories except for self-injurious and ritualistic behaviors. In summary, based on the RBS-R, girls had a higher frequency of self-injurious and ritualistic endorsed behaviors, but boys had a larger repertoire of both behaviors. However, fewer girls in the sample demonstrated self-injurious

behaviors. That is, girls who were self-injurious demonstrated these behaviors at a high frequency, but these represented a smaller percentage of the sample.

Scores in the CRI were nearly identical between boys and girls. There was little variation in subtest scores measuring just right and repetitive behavior and mean scores between the genders varied by less than a mean of 2.

There were six domains included in the RBS-R rating form. The first subscale was a stereotyped behavior subscale. This included body rocking and swaying, locomotion (turning in circles), object usage (spinning objects), and sensory interests. The second subscale measured self-injurious behaviors including: hitting with self or object, pulling, scratching, or skin picking. The third subscale included compulsive behaviors. These included arranging objects, washing and cleaning, hoarding and saving items, and repeating routines. The fourth subscale measured ritualistic behavior, which refers to activities performed in a routine-like manner. These behaviors may occur during events like mealtimes, bathroom use, play, or social interactions. The fifth subscale was the sameness behavior subscale. Behaviors included resistance to change and insisting that things stay the same and do not change. The last subscale in the RBS-R measured restricted behaviors. Limited range of interests and fascination with movement or objects were included in this category. The six subscales measured the higher and lower-level order RRBs discussed by Turner (1999). Turner categorized higher-level behaviors as: insistence on sameness, object attachment, repetitive language, and circumscribed interests. It is interesting to note that the female cohort in this study scored frequently in several of these higher-level categories.

Girls engaged in all behavioral categories of the RBS-R. Specifically they scored most frequently in the subtest of insistence on sameness. Insistence on sameness was also where the

greatest discrepancy was found between the two groups (boys mean 8.32; girls mean 9.97). Similar to the boy cohort, girls demonstrated a frequent occurrence of many RRBs. In fact, girls engaged in many behaviors that boys did. Even though they demonstrated similar RRBs, girls' profiles are still not being captured during the evaluation process. It appears that there is a qualitative difference between RRBs that girls engaged in as opposed to boys; a finding that has been discussed by other researchers (Halladay et. al 2015). This indicates that RRBs are not an accurate way to identify ASD in girls. RRBs in boys are distinguishable to parents and therapists. Boys scored highest in stereotyped behaviors which, previous research supports, is a hallmark sign of RRBs in ASD. It is difficult to pinpoint RRBs in girls as they engaged in nearly all behaviors.

The gender bias in ASD testing is supported in several studies. Dworzynski, Ronald, Bolton, & Happé (2012) compared children aged 10-12 with an ASD diagnosis to others without a diagnosis regardless of their severity score on the Childhood Autism Spectrum Test (CAST). They found that girls who met diagnostic criteria displayed comorbid concerns such as low intellectual levels and behavioral difficulties suggesting a gender bias in testing. Kopp and Gillberg (1992) conducted a study on six high functioning girls with ASD. Researchers found that, though the girls had motor delays and social/communicative deficits, they had not received a diagnosis until after age six. Additionally these children had higher IQ scores than expected (between 60-100). They suggested a potential difference in ASD phenotypes between genders. More recently a study using data from the Autism and Developmental Disabilities Monitoring Network found that girls with an IQ higher than 70 and met diagnostic criteria were less likely to receive a diagnosis of ASD (Giarelli et al. 2010).

It was not anticipated that girls would produce such a broad variety of RRBs. Because they engaged in so many different types of RRB subgroups, it is difficult to determine which, if any, behavior is specific to the girls. It appears that as children age, they engage in different types of RRBs with respect to gender. This information contributes to the need for different methods of assessment when identifying girls with ASD. However, there are several researchers who report that gender differences are not distinguishable between children with ASD or at least do not make an appearance until a certain age. In fact, children exhibit several times of the same behaviors making them indistinguishable from one another. This finding supports previous research conducted by Zwaigenbaum et. al (2012) who measured sex differences in cognitive functioning between boys and girls from a high and low risk group. Their study, comparing high and low risk children with ASD based on cognitive abilities and ADOS severity scores, found that there were not significant effects of gender or diagnostic group by sex interaction. The gender differences in cognitive/adaptive skills and ASD severity between boys and girls younger than three with ASD were mirrored in a non-ASD group. That is to say in their study, boys with ASD engaged in gender different skills that were similar to boys without ASD. Additionally, they found that girls diagnosed with ASD from the higher risk sample were more likely to lose their diagnosis after age three as they no longer met diagnostic criteria.

Based on these findings, RRBs cannot be a unique gauge of identification of girls with ASD and is indicative of the need of more reliable diagnostic tools. Though this conclusion was not anticipated in this study, it is consistent with those of Van Winjngaarden-Cremers et al., (2014). They found that boys showed more repetitive and stereotyped behaviors than girls. Additionally, they found that girls demonstrated fewer RRBs with no gender differences on social behavior, concluding that more work was needed to draw away from the male gender bias.

Another study observed sex differences in a sample of 551 children with ASD and TD. Using measures of ASD severity through the ADOS and cognitive function through the Mullen and Vineland, researchers found that there were no significant effects of gender on symptoms of ASD, adaptive functioning, or communication skills. Additionally, children with ASD (girls= 54, boys= 234) engaged in similar frequencies of RRBs as measured by the ADOS (girls= 4.04, boys= 3.72) (Reinhardt, Wetherby, Schatschneider, Lord 2014).

Literature supports the claim that boys are more involved in restricted interests. A study in 2007 DeLoache, Simcock, & Macari found that boys were twice as likely to have intense interests. The authors suggest, “gender differences occur primarily for the most extreme level of interest, such that examining only those interests would reveal a preponderance of boys at any age” pg 1585) There is potentially a range of restricted interests in boys in general (those with TD and ASD) but this category did not distinguish boys from girls in our sample.

Limitations

One limitation of this study is the reliability of parent report. Both measures of RRBS, the RSB-R and CRI, are dependent on caregiver accuracy. It is shown that these reports may be biased and are not always accurate. It is also worth noting the different reporting measures used in this study. The ADOS, an ASD severity rating scale, is a clinical measure, while the RBS-R and CRI are parent reported measures. There may be some discrepancy when using two diverse methods of data collection.

Another limitation of this research is the unequal number of boys to girls included in the study. As boys made up the majority of the study, the reduced sample of girls may have impacted the results. Though, the imbalance sample size is a limitation of the study, it is important to add that the gender bias is representative of the ratio of boys to girls with ASD.

According to the CDC, ASD occurs significantly more often in boys compared to girls, with a ratio of 4:1 (Baio et al., 2018) respectively. The current study included a sample size of 174 participants (143 boys, 32 girls), a ratio of approximately 4.47 to 1.

Conclusion

The purpose of this paper was to examine and describe RRBs in a cohort of children with ASD. It is important to identify characteristics of females with ASD and design better diagnostic tools in order to create a phenotype of ASD specific to females. Though no observable RRB patterns were observed, a sub sample of toddlers revealed that a fewer girls engaged in lower order behavior behaviors than boys. RRBs did not distinguish gender in this study suggesting that it is not a unique diagnostic indicator for girls. However, the RBS-R captured the rate and repertoire of RRBs in girls making it a valuable asset in distinguishing ASD from other diagnoses.

Table 1. Means and standard deviations of RRB measures including 6 subtests of RBS-R and CRI scores according to gender

<i>RBS-R Item</i>	Girls (<i>n</i> =32)			Boys (<i>n</i> =143)		
	Girls Mean	SD	Range	Boys Mean	SD	Range
RBS-R Stereotyped Endorsed	3.77	2.16	1-6	3.52	1.86	1-6
RBS-R Stereotyped Total	9.13	8.27	0-41	6.24	4.43	0-18
RBS-R Self Injurious Endorsed	2.26	2.44	0-8	2.49	2.52	0-8
RBS-R Self Injurious Total	4.68	6.24	0-23	4.18	5.44	0-24
RBS-R Compulsive Endorsed	4.10	2.52	0-9	3.46	2.36	0-9
RBS-R Compulsive Total	7.06	5.56	0-24	6.31	5.43	0-24
RBS-R Ritualistic Endorsed	2.52	2.23	0-6	2.63	2.04	0-6
RBS-R Ritualistic Total	5.10	5.72	0-18	5.03	4.84	0-18
RBS-R Sameness Endorsed	4.97	3.24	0-11	4.60	3.53	0-11
RBS-R Sameness Total	9.97	8.82	0-33	8.32	8.13	0-33
RBS-R Restricted Endorsed	2.32	1.58	0-4	2.53	1.41	0-4
RBS-R Restricted Total	4.71	4.05	0-12	5.01	3.86	0-12
RBS-R Endorsed Total	20.06	10.97	1-42	19.13	11.36	1-42
RBS-R Overall Total	41.65	31.34	1-124	34.96	26.99	1-124
<i>CRI Item</i>						

CRI- Just Right	12.67	6.35	5-25	12.65	5.85	4-24
CRI- Repetitive Behavior	11.27	4.74	4-20	11.05	4.98	4-20
CRI- IT	50.43	21.17	21-94	48.53	19.82	13-93

Table 2. Percentages of toddler sample that endorsed RRBs based on total scores

Type of RRB	Girls (n=19)	Boys (n=70)	Type of RRB according to Turner
Stereotyped	84%	93%	Lower
Self-Injurious	10%	71%	Lower
Compulsive	95%	86%	Higher
Ritualistic	74%	80%	Higher
Sameness	95%	89%	Higher
Restricted	79%	89%	Lower

Table 3. Means and standard deviations of toddler sample that endorsed RRBs based on total scores

Type of RRB	Girls		Boys	
	Mean	SD	Mean	SD
Stereotyped (total)	7.58	5.66	5.87	4.48
Self-Injurious (total)	4.12	6.01	3.97	5.27
Compulsive (total)	6.95	4.43	5.59	4.9
Ritualistic (total)	4.58	5.38	3.36	3.5
Sameness (total)	8.53	8.39	5.51	5.53
Restricted (total)	4.21	3.68	4.71	3.81

Table 4. Correlation Table

	RBS-R Overall Total	RBS-R Endorsed	ADOS SA Total	CRIt	Global Functioning
RBS-R Overall Total	1.00	0.50	-0.11	0.40	0.06
RBS-R Endorsed Total	0.50	1.00	-0.13	0.16	0.14
ADOS SA Total	-0.11	-0.13	1.00	-0.12	-0.28
CRIt	0.40	0.16	-0.12	-1.00	-0.02
Global Functioning	0.06	0.14	-0.28	-0.02	1.00

Table 5. Fixed Effects

	Estimate	Std. Error	df	t-value	PR (> t)
Intercept	1.69E+01	8.94E+02	1.62E+02	1.888	0.0608
Age	6.65E-06	1.17E-03	1.62E+02	0.006	0.9955
Gender	1.01E+00	4.19E+00	1.62E+02	0.242	0.8089
ADOS SA TOTAL	-2.29E-01	3.54E-01	1.62E+02	-0.649	0.5176
CRIt	5.12E-01	9.52E-02	1.62E+02	5.383	2.53e-07
Global Functioning	5.856e-02	7.590e-02	1.623e+02	0.772	0.4415

Figure 2. RBS-R Endorsed Item by Gender

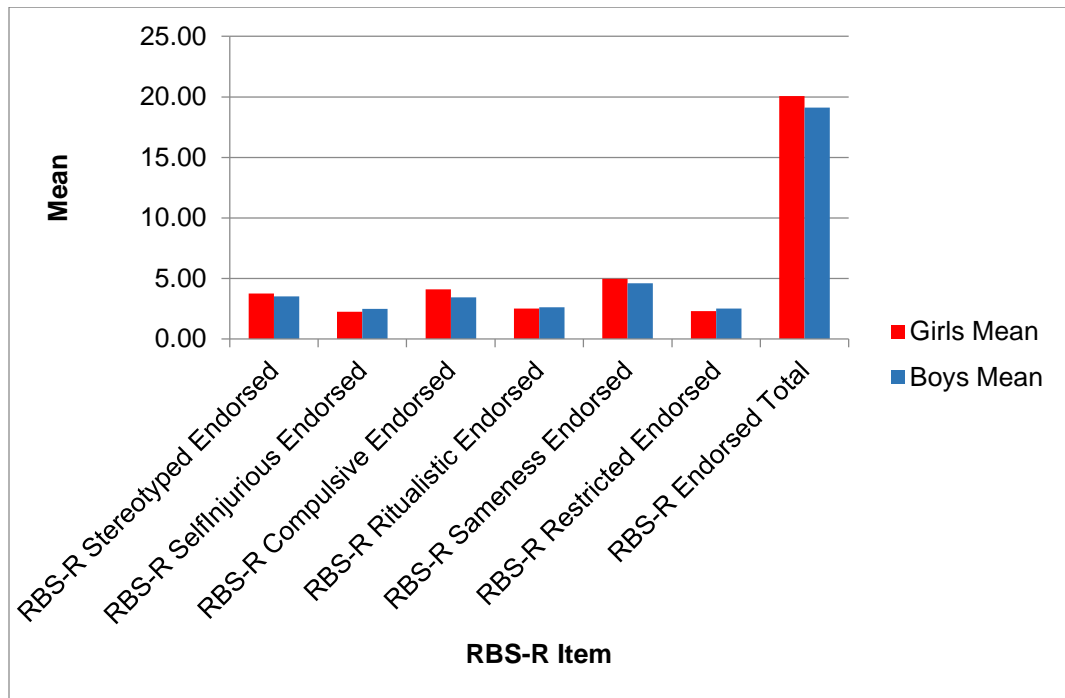


Figure 3. RBS-R Totals by Gender

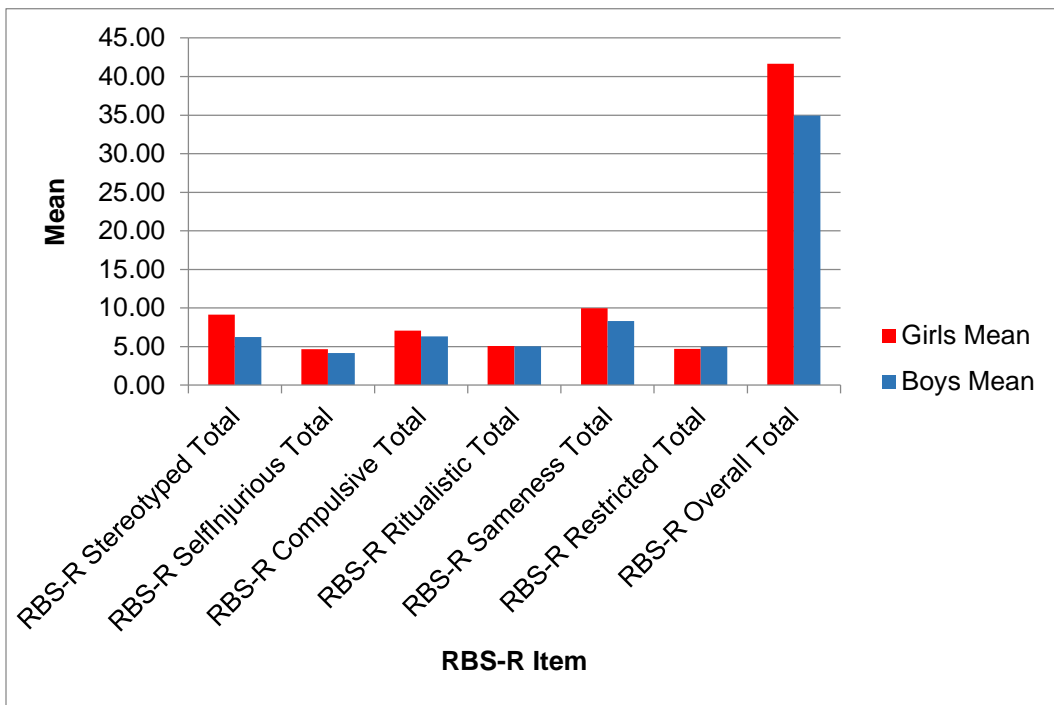
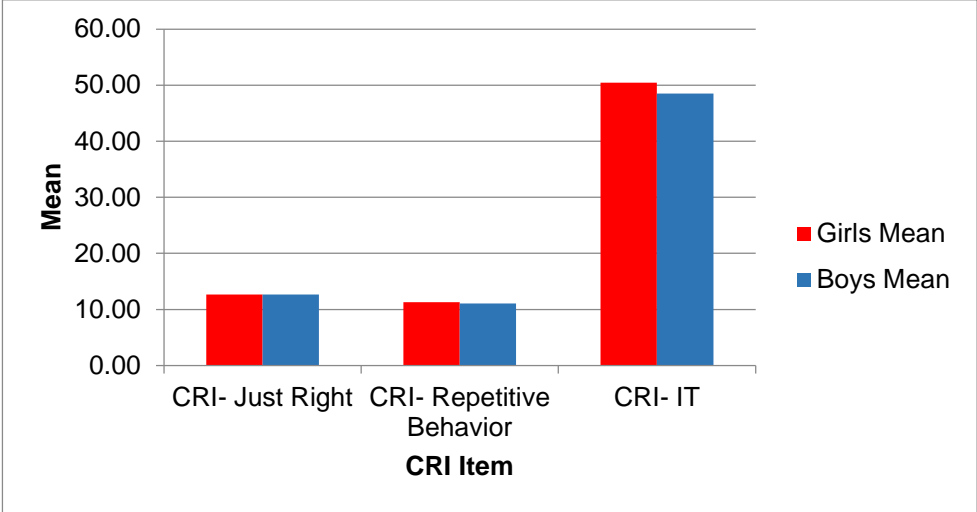


Figure 4. CRI totals according to gender



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APPENDIX



January 11, 2018

Theodore Tomeny, Ph.D.
Assistant Professor
Department of Psychology
College of Arts & Sciences
The University of Alabama
Box 870348

Re: IRB # 06-OR-022-R12 "Autism Spectrum Disorders Clinic Research Database"

Dear Dr. Tomeny:

The University of Alabama Institutional Review Board has granted approval for your proposed research. Your renewal application has been given expedited approval according to 45 CFR part 46. Approval has been given under expedited review category 7 as outlined below:

(7) Research on individual or group characteristics or behavior (including, but not limited to, research on perception, cognition, motivation, identity, language, communication, cultural beliefs or practices, and social behavior) or research employing survey, interview, oral history, focus group, program evaluation, human factors evaluation, or quality assurance methodologies.

Your application will expire on January 10, 2019. If your research will continue beyond this date, complete the relevant portions of the IRB Renewal Application. If you wish to modify the application, complete the Modification of an Approved Protocol Form. Changes in this study cannot be initiated without IRB approval, except when necessary to eliminate apparent immediate hazards to participants. When the study closes, complete the appropriate portions of the IRB Study Closure Form.

Please use reproductions of the IRB approved stamped consent/assent forms to obtain consent from your participants.

Should you need to submit any further correspondence regarding this proposal, please include the above application number.

Good luck with your research.

Sincerely,



Director & Research Compliance Officer
Office of Research Compliance

358 Rose Administration Building | Box 870127 | Tuscaloosa, AL 35487-0127
205-348-8461 | Fax 205-348-7189 | Toll free 1-877-820-3066