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Factors Associated with Restricted, Repetitive Behaviors and Interests and Diagnostic Severity Level in Young Children with Autism Spectrum Disorder

Esther Hong

Louisiana State University and Agricultural and Mechanical College

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**FACTORS ASSOCIATED WITH RESTRICTED, REPETITIVE
BEHAVIORS AND INTERESTS AND DIAGNOSTIC SEVERITY
LEVEL IN YOUNG CHILDREN WITH AUTISM SPECTRUM
DISORDER**

A Thesis

Submitted to the Graduate Faculty of the
Louisiana State University and
Agricultural and Mechanical College
in partial fulfillment of the
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Master of Arts

in

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by
Esther Hong
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Abstract

Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterized by deficits in social communication and social interactions and the presence of restricted, repetitive patterns of behavior (RRBIs). The presence of RRBIs can be detrimental to a child's development, as RRBIs can lead to impairments in other areas of functioning, impede learning, and contribute to parental stress. Previous studies have identified several factors that are associated with RRBIs severity and topography. The current study aims to assess whether impairments in adaptive functioning predict RRBIs severity, using the *Vineland Adaptive Behavior Scales, Third Edition (VABS-3)* and *Baby and Infant Screen for Children with Autism Traits (BISCUIT)-Part 1*, RRBIs subscale score. Additionally, clinician-assigned severity levels of ASD, *BISCUIT-Part 1*, *Childhood Autism Rating Scale, Second Edition (CARS2)*, and *Vineland VABS-3*, will be used to examine factors associated with ASD severity level. The findings of this study will provide implications for the early assessment and treatment of RRBIs in young children with ASD.

Introduction

Individuals with autism spectrum disorder (ASD) exhibit impairments in social communication and social interaction as well as restricted, repetitive behaviors, interests, and activities (American Psychological Association [APA], 2013). According to a recent report by the Centers for Disease Control and Prevention (CDC), ASD now affects 1 in 59 children in the United States (Baio et al., 2018). With increased prevalence and awareness of ASD, there has been an emphasis on the early detection and treatment of the core symptoms of ASD. Although early intensive behavioral intervention (EIBI) has demonstrated significant improvements in the areas of socialization, cognition, and language in children with ASD (Landa, 2018; Zwaigenbaum et al., 2015), RRBI have not been a primary focus of early interventions (Leekam, Prior, & Uljarevic, 2011; Odom et al., 2010).

If RRBI are not effectively treated, they can significantly impair daily functioning and result in poor, long-term outcomes (Koegel & Covert, 1972; Pierce & Courchesne, 2001; Raulston & Machalicek, 2018). The wide range of topographies and subtle changes in RRBI over time have made the assessment and monitoring of RRBI challenging. In response to these challenges, researchers have developed indirect (e.g., Bodfish, Symons, & Lewis, 1999; Le Couteur, Lord, & Rutter, 2003) and direct methods of assessment (e.g., Lord et al., 2012) for the early detection of RRBI in young children who are at risk for ASD. Additionally, researchers have examined the associations between various individual-specific factors (e.g., ASD symptom severity, age, intellectual functioning, gender, adaptive functioning) and the presentation of RRBI.

The present study aims to examine the relationship between ASD severity and RRBI severity in infants and toddlers with ASD. Specifically, this study will evaluate if the severity

level for ASD (i.e., Level 1, Level 2, Level 3) according to the *Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5; APA, 2013)* is associated with parent-reported severity ratings of RRBI. Additionally, the current study aims to address the gap in the existing literature by evaluating the associations between adaptive functioning and RRBI. The findings of this study will expand on the existing literature by examining how various factors influence the presentation of RRBI in infants and toddlers with ASD.

Autism Spectrum Disorder

History of ASD

The first account of autism as it is understood today was described by Leo Kanner in 1943. Kanner described eleven cases (i.e., 8 males, 3 females) of young children who presented with a “unique syndrome” that differed from childhood schizophrenia. While the children displayed individual differences in the degree of their impairments and the manifestation of symptoms, Kanner detailed several core symptoms that were exhibited by all children. Notably, he reported that all eleven children displayed an “extreme autistic aloneness” starting at infancy, in which they had the inability to relate themselves to people and situations (Kanner, 1943). Other symptoms included the delay in speech, “excellent” rote memory, atypical speech (i.e., echolalia, pronoun reversal, literal use of language), repetitive behaviors (i.e., noises, motions, activities), insistence on sameness, limited spontaneous activity, and sensitivity to food, loud noises, and moving objects. In a follow-up study, Kanner (1971) noted that although the developmental trajectory of the symptoms differed across children, the children’s language, socialization, and challenging behaviors (e.g., tantrums, food and noise sensitivities) appeared to improve with age while IQ decreased with age.

Kanner’s study (1943) also provided a theory for the cause of infantile autism. Kanner described the children’s parents as highly intelligent individuals who were cold, formal, obsessive, and uninterested in people. He hypothesized that the parents’ behavior contributed to the children’s autistic symptoms. Consequently, parents of children with autism, particularly the mothers, were blamed for their child’s autistic symptoms (Bettelheim, 1967). In a subsequent study, Rutter (1968) challenged Kanner’s theory, arguing that higher rates of autism would be found in the siblings of children with autism if parenting styles did, in fact, cause autism.

One year after Kanner's seminal publication, Hans Asperger published his thesis, in which he described 4 children with a disorder he coined, "autistic psychopathology" (Asperger, 1944). Although Asperger's work did not receive attention until it was translated into English by Lorna Wing in 1981, there were many similarities between the two authors' accounts of autistic behavior. Similar to Kanner's cases, the children described in Asperger's study demonstrated social withdrawal, impairments in the development of social and emotional relationships, idiosyncratic language, and RRBI's (e.g., stereotypic behaviors, restricted interests). These symptoms presented in early childhood and persisted throughout the lifespan. However, the children in Asperger's study demonstrated savant-like skills in mathematics or natural sciences (1944). Asperger also highlighted similar personality traits between the parents and children, suggesting that autistic psychopathology may be an "extreme variant" of intelligence (Wolff, 2004).

Both Kanner and Asperger emphasized the distinction between autism and childhood schizophrenia. Although both authors used the term "autistic" to describe the core feature of infantile autism and autistic psychopathology, the use of this term differed from its original use by Bleuler (1911). Bleuler first coined the term, "autistic," to describe characteristics exhibited by individuals with schizophrenia. However, Kanner stated that infantile autism was distinguishable from childhood schizophrenia because children with infantile autism exhibited autistic aloneness starting at the beginning of life, whereas children with schizophrenia exhibited a departure from previously established relationships (Kanner, 1943). Similarly, Asperger distinguished autistic psychopathology from schizophrenia, such that individuals with autistic psychopathology had a life-long, stable personality while individuals with schizophrenia demonstrated a progressive change in personality (Asperger, 1944).

In the translated publication of Asperger's thesis, Wing added to Asperger's original account of autistic psychopathology and coined the term, "Asperger's syndrome," as it is conceptualized today (1981). She is credited with expanding the diagnostic criteria of autism and describing the disorder as a spectrum of autistic disorders (Hippler & Klicpera, 2003). Further, she characterized Asperger's syndrome as a triad of impairments in socialization, communication, and imagination.

Diagnostic Criteria

Although infantile autism was first described by Kanner in 1943, formal diagnostic criteria for autism was not published until 1980, in the APA's *Diagnostic and Statistical Manual of Mental Disorders, Third Edition (DSM-III)*. Under the category of pervasive developmental disorders (PDD), three separate diagnoses were included: infantile autism, childhood onset PDD, and atypical PDD. Differential diagnosis between these three PDDs were made primarily on the onset of the disorder and the range of impairment. Infantile autism specified the age of onset prior to 30 months of age while childhood onset PDD specified the age of onset between 30 months and 12 years of age. Atypical PDD was used to describe children with several developmental deficits in socialization and language but did not meet criteria for infantile autism or childhood onset PDD (Volkmar, Cohen, & Paul, 1986). Although there were no objective assessment measures for infantile autism at the time of publication, the *DSM-III* criteria were the first to provide explicit descriptive criteria for the diagnosis of autism.

Several years later, in the *Diagnostic and Statistical Manual of Mental Disorders, Third Edition, Revised (DSM-III-R; APA, 1987)*, the diagnostic criteria for autism underwent several changes. The diagnostic label of infantile autism was changed to Autistic Disorder and the 2 PDDs were re-classified as Pervasive Developmental Disorder-Not Otherwise Specified (PDD-

NOS). Autistic Disorder was characterized by a triad of impairments, in (1) reciprocal social interaction, (2) verbal and nonverbal communication, and (3) restricted activities and interests. The diagnosis of PDD-NOS was assigned to those with qualitative impairments in socialization and communication but did not meet the full criteria for Autistic Disorder. While the age of onset for Autistic Disorder was not specified, the diagnostic criteria did indicate an onset during infancy or early childhood.

The diagnostic criteria for autism was expanded in the *Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition (DSM-IV; APA, 1994)* to include 5 distinct PDDs: Autistic Disorder, Asperger's Disorder, PDD-NOS, Rett's Disorder, and Childhood Disintegrative Disorder. The triad criteria for Autistic Disorder was maintained but was revised to (1) impairments in social interaction, (2) impairments in communication, and (3) restricted, repetitive, and stereotyped patterns of behaviors, interests, and activities. In order to qualify for Autistic Disorder, impairments in at least one of the three domains must have onset prior to 3 years of age. The diagnosis of Asperger's Disorder was assigned to individuals with impairments in socialization and restricted, repetitive behaviors but no impairments in cognition, language, or adaptive functioning. The diagnosis of PDD-NOS was assigned to individuals who had impairments in social interaction, communication, and stereotyped behaviors but had a late age onset or did not meet full diagnostic criteria for Autistic Disorder. Rett's Disorder specified the development of deficits (i.e., in social engagement, motor skills, and language) between 5 and 48 months of age, following normal development. The diagnostic criteria for Childhood Disintegrative Disorder also involved the regression of skills following a period of at least 2 years of apparently normal development.

In 2013, the APA published the current diagnostic criteria for ASD in the *DSM-5*. This revision aimed to address the challenges of categorizing the heterogeneous presentations of ASDs into 5 distinct subgroups of ASD (Grzadzinski, Huerta, & Lord, 2013). Therefore, the *DSM-5* replaced the *DSM-IV*'s multi-categorical method with one diagnostic category of ASD. Consequently, Autistic Disorder, Asperger's Disorder, and PDD-NOS were subsumed under one diagnosis of ASD, and Rett's Disorder and Childhood Disintegrative Disorder were removed from the *DSM-5*. In addition to this change, several more changes were introduced. First, the *DSM-5* moved away from the triad model of impairments, to a dyad model. The new diagnostic criteria for ASD is characterized by (1) deficits in social communication and social interaction and (2) restricted, repetitive patterns of behaviors, interest, or activities. The *DSM-IV* requirement of impairments in communication was removed to reflect the research that impairments in communication were not specific to individuals with ASD (Hartley & Sikora, 2006; Matson & Neal, 2010). In line with the existing research that sensory behaviors are commonly found in individuals with ASD (Ben-Sasson et al., 2009; Leekam, Nieto, Libby, Wing, & Gould, 2007), "hyper- or hypo-reactivity to sensory input or unusual interests in sensory aspects of the environment" was added as a distinct RRBI symptom. To aid with the differential diagnosis of comorbid conditions, specifiers (i.e., with or without accompanying intellectual impairment; with or without accompanying language impairment; associated with a known medical or genetic condition or environmental factor; associated with another neurodevelopmental, mental, or behavioral disorder; with catatonia) were also introduced. Lastly, the diagnostic criteria for ASD now includes an ASD severity rating for each domain, which will be discussed in detail in the following section.

Severity Level for ASD

With the *DSM-5*, a dimensional assessment of ASD severity was introduced. This change to the diagnostic criteria allows clinicians to assign a severity level to each of the two core features of ASD (i.e., social communication and RRBI; APA, 2013; Mazurek, Lu, Macklin, & Handen, 2018). A severity rating of “Level 1” indicates that the child requires support, “Level 2” indicates that the child requires substantial support, and “Level 3” indicates that the child requires very substantial support. Although the *DSM-5* provides some qualitative guidance for clinicians and researchers, there are no objective methods on how severity ratings should be determined (Mechling & Tassé, 2016). For instance, it is not clear if clinicians and researchers are assigning ASD severity level according to the severity of the core symptoms of ASD or if they are making determinations strictly based on the child’s need for support and intervention. In the latter case, it is unclear if other areas of impairment (e.g., cognition, language, challenging behaviors) contribute to the level of support required (Mazurek et al., 2018). Thus, it is likely that clinicians and researchers are conceptualizing severity levels subjectively.

Although this area of research has been understudied, researchers have recently begun to evaluate factors that contribute to determinations of ASD severity level. In regard to the level of functional impairment and ASD severity level, Weitlauf et al. (2014) reported mixed associations between a child’s impairment across domains (i.e., cognitive, adaptive, ASD-specific symptoms) and assigned ASD severity level, which suggests that there is no uniform method of assigning ASD severity level based on a child’s level of impairment. Nevertheless, there appears to be some consistency among parental ratings of severity, clinician ratings of severity, and behavioral observations. In a recent study, Mazurek et al. (2018) found consistency between parent-reported RRBI severity scores (i.e., according to the *Aberrant Behavior Checklist*; Aman &

Singh, 1986), diagnostic observation score (i.e., according to the *ADOS-2*; Lord et al., 2012), and ASD severity level. However, there was no association between parental ratings and clinician ratings of severity on the social communication domain, as the parental rating of social withdrawal was not associated with ASD severity rating. Further, intellectual functioning and age were found to influence ASD severity ratings on both the social communication and RRBI domains (Mazurek et al., 2018), which indicates that other areas factors are conflating ASD severity.

Prevalence of ASD

Autism was once an uncommon disorder, with prevalence rates of approximately 4-5 children per 10,000 (Howlin, 2006; Rutter, 1968). However, prevalence rates have steadily increased since these early reports. When the CDC first began monitoring the prevalence of ASD in children in the United States, the prevalence was approximately 1 in 150 children (CDC, 2007). Subsequent studies by the CDC reported an increase in the prevalence, from 1 in 88 children in 2012, to 1 in 59 children in 2018 (Baio et al., 2018). While there are no definitive reasons for the dramatic increase in prevalence rates, it appears that there are several factors that may account for this increase. Some potential explanations include the expansion of the diagnostic criteria, increased awareness of ASD, early assessment, cultural factors, environmental factors, and improvements in research methodology (Fombonne, 2009; Matson & Kozlowski, 2011).

Early Assessment of ASD

As the awareness of ASD has risen, there has been an increased emphasis on the early identification of ASD. In response, researchers have designed screening tools (Robins, Fein, & Barton, 1999; Matson, Boisjoli & Wilkins, 2007) and observation scales (Gotham, Risi, Pickles

& Lord, 2007; Schopler, Van Bourgondien, Wellman, & Love, 2010) for the early screening and diagnosis of ASD in toddlers under 3 years of age. Additionally, there has been increased research on parental age of first concern, as parents of children with ASD report developmental concerns as early as 12 months of age (Matheis et al., 2017; Ozonoff et al., 2009). Although the existing literature supports that ASD can be reliably diagnosed in toddlers as young as 18 months of age (Chawarska et al., 2014; Daniels & Mandell, 2014; Kuban et al., 2009), the majority of children do not receive an ASD diagnosis before the age of 5 years (Shattuck et al., 2009). Indeed, with growing evidence for the efficacy of EIBI to improve long-term outcomes for children with ASD (Landa, 2018; Reichow, Barton, Boyd, & Hume, 2012; Virués-Ortega, 2010), the early assessment of ASD is critical in order to mitigate the impairments associated with ASD.

Restricted, Repetitive Behaviors and Interests (RRBIs)

Although RRBIs are found among infants with typical development as well as children with other developmental delays, children with ASD consistently exhibit higher rates and a wider range of topographies of RRBIs in comparison to other groups (Bodfish, Symons, Parker, & Lewis, 2000; Harrop et al., 2014; Kim & Lord, 2010; Watt, Wetherby, Barber, & Morgan, 2008). According to the *DSM-5* diagnostic criteria, there are four categories of RRBIs: (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, (4) hyper- or hypo-reactivity to sensory input or unusual interest in sensory aspects of the environment (APA, 2013).

In the existing literature, RRBIs are generally categorized into two subtypes: low-level RRBIs and high-level RRBIs (Leekam, Prior, & Uljarevic, 2011; Raulston & Machalicek, 2018; Turner, 1999). This categorization is in reference to the functioning level of the children that typically display a specific topography of RRBI. That is, that low-level RRBIs have been observed in children of younger age, greater developmental delays, and lower cognitive ability (Prior & Macmillan, 1973; Turner, 1999), whereas high-level RRBIs have been observed in children with higher cognitive and language abilities (Bishop, Richler, & Lord, 2006; Esbensen, Seltzer, Lam, & Bodfish, 2009; Richler, Huerta, Bishop, & Lord, 2010). Low-level RRBIs include behaviors such as stereotyped, repetitive motor movements (e.g., hand flapping, body rocking), object use (e.g., lining up objects), and sensory behaviors (Rapp & Vollmer, 2005; South, Ozonoff, & McMahon, 2005). High-level RRBIs include perseverative interests, repetitive questioning, obsessions, and compulsions (Boyd, McDonough, & Bodfish, 2012).

These distinctions in the presentation of RRBI has led researchers to evaluate additional factors associated with the presentation of RRBI.

Assessment of RRBI

Given that the presence of RRBI is a core feature of ASD, they manifest early in a child's development and can be detected as early as 17-37 months of age (Matson, Dempsey, & Fodstad, 2009; Ozonoff et al., 2008; Rogers, 2009; Yirmiya & Charman, 2010). Indeed, the presence of RRBI during early childhood is one of the most reliable predictors of a future ASD diagnosis (Lord & Luyster, 2006; Lord et al., 2006). However, the assessment of RRBI can be challenging, as RRBI are behaviorally-defined symptoms that can take many forms (Bodfish et al., 2000; Lewis & Bodfish, 1998). The assessment process is further complicated by the overlap in RRBI presentation. For example, repetitive tapping of the ears may be classified as a stereotyped motor behavior or as a sensory-seeking behavior (APA, 2013). Furthermore, there is a lack of sensitive screening and assessment measures that can detect the subtle differences in RRBI and changes in RRBI over time (Honey, Rodgers, & McConachie, 2012; Raultson & Machalicek, 2018). This has detrimental consequences for the early assessment of RRBI as well as for treatment monitoring.

In response to this gap in research, researchers have developed several measurement tools for the screening and assessment of RRBI. While there has been a recent increase in the use of standardized, direct observational methods to assess RRBI (Lord et al., 2012), the most commonly used methods of assessment are indirect methods, such as questionnaires (e.g., *Repetitive Behaviour Scale*; Bodfish et al., 1999), rating scales, and interviews (e.g., *Autism Diagnostic Interview- Revised*; Le Couteur et al., 2003) with parents and caregivers (Honey et al., 2012; Kim & Lord, 2010; Lewis & Bodfish, 1998). These methods of informant-based

measures have been supported in the research, as parents have demonstrated the ability to reliably identify ASD symptoms in their children as early as 12-18 months of age (Gray & Tonge, 2005; Reznick, Baranek, Reavis, Watson, & Crais, 2007). However, there are limitations to these existing measures that must be taken into consideration. Although these existing assessment tools measure a wide range of RRBI commonly exhibited by individuals with ASD, there is a lack of research support for their use with subcategories of RRBI (Honey et al., 2012). Additionally, there are inconsistencies in how RRBI are categorized (Hus, Pickles, Cook, Risi, & Lord, 2007) and quantified (e.g., frequency, severity, nature) across measures (Honey et al., 2012).

Although research on the assessment of RRBI has expanded considerably over the past couple decades, improvements in measurement tools are still needed in order to better understand the nature of RRBI in individuals with ASD. In particular, investigating differences in RRBI among individuals with ASD may help to differentiate subgroups of children with ASD according to RRBI presentation (Honey et al., 2012). This type of fine-grained assessment of RRBI will help to guide individualized intervention plans and monitor subtle changes in RRBI throughout the course of treatment.

Prognosis and Treatment of RRBI

With the growing research support for early intervention, there has been an increased focus on the early treatment of infants and toddlers with ASD (Chawarska, Klin, Paul, & Volkmar, 2007). While EIBI has demonstrated marked improvements in various skill areas such as socialization, cognition, and language in children with ASD (Landa, 2018; Zwaigenbaum et al., 2015), there has been a dearth of studies evaluating the effect of EIBI on RRBI (Leekam, Prior, & Uljarevic, 2011; Lewis & Bodfish, 1998; MacDonald et al., 2007) or the role of RRBI

as predictors of outcome (Troyb et al., 2016). Even though stereotyped behaviors are the most commonly reported challenging behavior among children receiving EIBI services (Hong et al., 2018; Jang, Dixon, Tarbox, & Granpeesheh, 2011), RRBI are not a primary target of most comprehensive behavioral interventions (Odom, Boyd, Hall, & Hume, 2010). When they are, there are less significant improvements in RRBI in comparison to other core symptoms of ASD (Fecteau, Mottron, Berthiaume, & Burack, 2003). This is extremely concerning given that RRBI can cause impairments across several areas of functioning (e.g., social, self-help, language), impede acquisition of new skills (Koegel & Covert, 1972; Pierce & Courchesne, 2001; Raulston & Machalicek, 2018), and contribute to greater parental stress (Hayes & Watson, 2013). Additionally, if early intervention for RRBI is not provided, RRBI can become entrenched and difficult to change (Leekam, Prior, & Uljarevic, 2011).

The treatment of RRBI is complex, as the severity, frequency, and topography of RRBI can change across the lifespan (Johnson, McConachie, Watson, Freeston, & Le Couteur, 2006; Lam & Aman, 2007). Treatment of RRBI is further complicated because RRBI are usually maintained by automatic or nonsocial reinforcement, which means that they are less susceptible to behavior change (Raulston & Machalicek, 2018). Nevertheless, emerging research suggests that behavior-based interventions, such as antecedent modifications and integration of RRBI (e.g., fixated interests, preoccupation with objects) as reinforcers in treatment have been effective in reducing certain topographies of RRBI (Boyd, Conroy, Mancil, Nakao, & Alter, 2007; Kryzak & Jones, 2015; Charlop-Christy & Haymes, 1996; Vismara & Lyons, 2007). Previous research also indicates that there are several individual-specific variables (e.g., age, IQ, language skills, adaptive functioning) that are associated with RRBI. However, more longitudinal studies

evaluating the developmental trajectories of RRBI and treatment effects on RRBI are needed to understand how RRBI change over time.

RRBI in Children with ASD

Most of the extant research on the differential presentation of RRBI are comparison studies of children with ASD, children with other developmental delays, and children with typical development. As a result, it is unclear how different RRBI manifest among children with ASD and which factors are associated with the changes in RRBI. It is incumbent upon researchers to investigate the subtle differences in RRBI, specifically in individuals with ASD, in order to develop more sensitive screening tools, improve early assessment, and guide treatment planning (Raulston & Machalicek, 2018). Expanding research in this area will help clinicians and researchers understand which RRBI change with age, which RRBI are more resistant to intervention, which RRBI are more impairing and impede acquisition of skills, which settings and contexts RRBI are more prevalent in, which RRBI warrant comprehensive intervention versus focused interventions, and so on.

Factors Associated with RRBI

The prevalence and presentation of RRBI vary considerably depending on individual differences such as age, intellectual functioning, gender, and adaptive functioning (Bradley, Boan, Cohen, Charles, & Carpenter, 2016; Leekam, Prior, & Uljarevic, 2011). These factors associated with RRBI are described in detail below.

Age

There are mixed findings in regard to how RRBI in individuals with ASD change over time. Several studies have found that the severity of RRBI is the highest during early childhood but decreases with age (Esbensen et al., 2009; South et al., 2005). However, this trajectory has not been found during early childhood, as RRBI (e.g., repetitive use of objects, atypical hand and finger movements, unusual preoccupations, compulsions and rituals) were exhibited at higher rates by children aged 4-5 years than children aged 2-3 years (MacDonald et al., 2007; Moore & Goodson, 2003).

The relationship between RRBI and age appears to be dependent on the behavior in question. For instance, Murphy et al. (2005) found that the frequency of atypical motor movements and sensory behaviors reduced with age while the frequency of resistance to change and adherence to routines behaviors did not. Additionally, repetitive use of objects has been found to decrease with age (Fecteau et al., 2003) and restricted interests has been found to increase with age (South et al., 2005; Richler et al., 2010).

In a longitudinal study of young children with ASD, repetitive motor and sensory behaviors occurred at high rates across age 2, 3, 5, and 9 years (Richler et al., 2010). Similar to the findings in the aforementioned studies, these RRBI occurred at significantly lower rates at 9 years of age. However, this trend was only observed among children with higher nonverbal IQ,

which suggests that other factors, such as intellectual ability, influence the developmental trajectory of RRBI. Richler et al. (2010) also found that insistence on sameness (i.e., routines and rituals) appeared at 2 years of age and moderately increased in severity with age, demonstrating that insistence on sameness behaviors increase with age. These findings support the claim that low-level RRBI are more commonly found in younger children and tend to decrease with age, while high-level RRBI increase with age (Cuccaro et al., 2003; Richler, Bishop, Kleinke, & Lord, 2007).

Intellectual Functioning

Approximately 70% of individuals with ASD have intellectual disability (ID; Mannion, Leader, & Healy, 2013; Matson & Nebel-Schwalm, 2007). As a result, intellectual functioning has been highly studied in ASD research. Individuals with ASD exhibit the highest rates of stereotyped behavior in comparison to individuals with ID only or other comorbid conditions (Esbensen et al., 2009; Rojahn, Matlock, & Tassé, 2000). When intelligence is considered, individuals with ASD and lower intelligence quotient (IQ) evince greater RRBI severity than individuals with ASD and higher IQ (Mazurek, 2018). This association is also found in young children with ASD, such that toddlers with greater impairment in developmental functioning showed higher severity of RRBI (Matson et al., 2013). Intellectual functioning also appears to influence the topography of RRBI, such that children with higher intelligence exhibit high-level RRBI and children with lower intelligence exhibit low-level RRBI (Lam & Aman, 2007; Hus et al., 2007; South et al., 2005). Despite the abundance of research on this topic, the role of intelligence on RRBI remains unclear. That is, does intelligence conflate RRBI severity or is it a distinct variable that is associated with RRBI?

ASD Severity

There is a strong relationship between the core symptoms of ASD (Dworzynski, Happé, Bolton, & Ronald, 2009; Kuenssberg & McKenzie, 2011), which suggests that overall severity of ASD is correlated with impairment in RRBI. Using the *DSM-IV* diagnostic categories of ASD, Matson et al. (2009) investigated the relationship between ASD severity and RRBI in toddlers with Autistic Disorder and PDD-NOS. The authors reported that toddlers with a diagnosis of Autistic Disorder exhibited a greater number of RRBI than toddlers with a diagnosis of PDD-NOS. Further, children with Autistic Disorder had higher severity ratings on all 30 RRBI factor items on the *BISCUIT-Part 1*, Repetitive Behavior/Restricted Interest subscale.

To date, only one study (Mazurek et al., 2018) has evaluated the relationship between RRBI and ASD severity levels according to *DSM-5* criteria. The results from this study were reported in the previous “Severity Levels for ASD” section. No other studies have conducted an exploratory analysis of differences in RRBI presentation according to ASD severity level.

Gender

Since the publication of Kanner’s study in 1943, there has been a large gender disparity in ASD. Currently, the male-to-female ratio in ASD is approximately 4:1 (Baio et al. 2018; Hill, Zuckerman, & Fombonne, 2016). Recently, researchers have started to explore the reasons for gender differences in the presentation of ASD symptoms, particularly in RRBI. Unfortunately, the findings in the existing literature are mixed. Several studies have found no significant gender differences in RRBI (Andersson, Gillberg, & Miniscalco, 2013; Banach et al., 2009; Carter et al., 2007; Lawson, Joshi, Barbaro, & Dissanayake, 2018). However, other studies have identified gender differences, such that females with ASD exhibit fewer RRBI than males with ASD (Hartley & Sikora, 2009; Frazier & Hardan, 2017; Sipes, Matson, Worley, & Kozlowski,

2011). Some researchers have also reported gender differences in the topography of RRBI. For instance, males with ASD have been found to demonstrate higher rates of repetitive use of objects, preoccupation with parts of objects, and adherence to rituals in comparison to females with ASD (Hiller, Young, & Weber, 2014; Matheis, Matson, Hong, & Cervantes, 2018; Nicholas et al. 2008). Further analyses on gender differences within the subcategories of RRBI is needed to improve the diagnostic process and understand how gender influences changes in RRBI across the lifespan.

Adaptive Functioning

Although individuals with ASD experience impairments in adaptive functioning, it is not a requirement in the diagnostic criteria, as it is for the ID diagnosis (APA, 2013). Nevertheless, many individuals with ASD experience pervasive impairments in adaptive functioning (Klin, Volkmar, & Sparrow, 1992; Volkmar, Lord, Bailey, Schultz, & Klin, 2004). Although adaptive functioning appears to covary with RRBI, this relationship has been overlooked in the existing literature (Cuccaro et al., 2003). Consequently, the relationship between adaptive functioning and RRBI in individuals with ASD remains unclear. Using a sample of individuals with ID with and without stereotypic behaviors, Matson, Kiely, and Bamburg (1997) found that participants with higher stereotypic behaviors had significantly lower scores on all 3 adaptive domains (i.e., communication, daily living skills, and socialization). In individuals with ASD, lower adaptive behavior composite scores (i.e., according to the VABS) were found be associated with higher repetitive motor and sensory behavior (Cuccaro et al., 2003). Further exploration of the relationship between adaptive functioning and RRBI may have significant implications for clinical practice (e.g., teaching adaptive skills to decrease RRBI, identifying subtypes in ASD).

Purpose

The presence of RRBI is a core diagnostic feature of ASD that can impede learning and lead to negative, long-term outcomes. Although researchers have evaluated various factors (e.g., age, IQ, gender, ASD severity) associated with the nature of RRBI, the relationship between ASD severity and RRBI still remains unclear, particularly in infants and toddlers with ASD. Moreover, the existing literature has not sufficiently examined associations between adaptive functioning and RRBI (Cuccaro et al., 2003). Previous studies have found that ASD severity ratings are positively correlated with parent-reported ratings of RRBI severity (Mazurek et al., 2018) and that adaptive functioning is negatively correlated with severity of RRBI (Cuccaro et al., 2003; Matson et al., 1997). These findings suggest that children with more severe ratings of ASD and deficits in adaptive functioning are likely to experience greater impairments in RRBI.

Therefore, the current study aims to examine the relationship among ASD symptoms, adaptive functioning, RRBI, and ASD severity. Although the existing literature provides evidence that these relationships exist, few studies have conducted a fine-grained analysis of these associations since the *DSM-5* diagnostic criteria for ASD was introduced in 2013. The current study will use the clinician-assigned ASD severity level, *Baby and Infant Screen for Children with Autism Traits - Part 1 (BISCUIT-Part 1)*, Restricted Behavior/Restricted Interests subscale, and the *Vineland Adaptive Behavior Scales, Third Edition (VABS-3)* to examine ASD severity level, RRBI severity, and adaptive functioning, respectively. Further, the *BISCUIT-Part 1*, Socialization/Nonverbal Communication subscale will be used as a measure of social skills delay, the *BISCUIT-Part 1*, the *BISCUIT-Part 1*, Communication subscale will be used as a measure of communication delay, and the *CARS2* will be used as a clinician-rated measure of ASD symptomatology and severity. Results from the present study may yield a detailed analysis

of RRBI frequency, severity, and topography in infants and toddlers with ASD and provide guidance for the early assessment and treatment of RRBI in children with ASD.

Method

Participants

Participants in the current sample were recruited through EarlySteps, Louisiana's statewide early intervention program. Under the Individuals with Disabilities Act, Part C, EarlySteps provides services to infants and toddlers under the age of 36 months, who have or are at risk for having a developmental delay. Children enrolled in EarlySteps who were found to be "at risk" for ASD according to an ASD screener (i.e., *Baby and Infant Screen for Children with Autism Traits, Part 1*; Matson et al., 2007) were referred to Louisiana State University's Psychological Services Center for a formal assessment of developmental functioning. The data for the current study were extracted from a pre-existing research database containing this assessment information.

To be included in the study, participants had to meet the following criteria: (a) were 37 months or under at the time of assessment, (b) have a DSM-5 diagnosis of ASD, (c) were administered the *BISCUIT- Part 1*, (d) were administered the *VABS-3*, and (e) were administered the *CARS2*. These criteria were applied to a pool of 317 children in the database, which resulted in a final sample size of 91 participants. The age of participants ranged from 19 to 37 months ($M = 30.08$, $SD = 4.31$). The study participants were 84.6% male ($n = 77$) and 15.4% female ($n = 14$). Of the total sample, 20.9% were African American, 64.8% were White, 4.4% were Hispanic, and 9.9% were identified as another ethnicity. Participant characteristics of the study sample are reported in Table 1. Participants were assigned to one of three groups based on their ASD severity level: ASD-Level 1, ASD-Level 2, and ASD-Level 3. The ASD diagnoses were given by a licensed clinical psychologist based on results from formal assessment measures, parent interview, and direct observation of the child in the clinic.

Table 1. Demographic information of the total sample and by group

| | Total (<i>N</i> = 91) | ASD-Level 1 (<i>n</i> = 6, 6.6%) | ASD-Level 2 (<i>n</i> = 30, 33%) | ASD-Level 3 (<i>n</i> = 55, 60.4%) |
|-------------------|---------------------------|--------------------------------------|--------------------------------------|--|
| Gender [N (%)] | | | | |
| Male | 77 (84.6%) | 6 (100%) | 24 (80%) | 47 (85.45%) |
| Female | 14 (15.4%) | 0 (0%) | 6 (20%) | 8 (14.55%) |
| Age in months | | | | |
| M (SD) | 30.08 (4.31) | 29.17 (2.99) | 29.80 (5.23) | 30.33 (3.91) |
| Range | 19 - 37 | 24 - 33 | 19 - 37 | 20 - 37 |
| Ethnicity [N (%)] | | | | |
| African American | 19 (20.9%) | 0 (0%) | 5 (16.67%) | 14 (25.45%) |
| White | 59 (64.8%) | 6 (100%) | 19 (63.33%) | 34 (61.82%) |
| Hispanic | 4 (4.4%) | 0 (0%) | 1 (3.33%) | 3 (5.45%) |
| Other | 9 (9.9%) | 0 (0%) | 5 (16.67%) | 4 (7.27%) |

Measures

The *Baby and Infant Screen for Children with aUtism Traits-Part 1 (BISCUIT-Part 1;* Matson, Boisjoli & Wilkins, 2007) is the diagnostic component of the *BISCUIT*, a three-part, informant-based assessment battery designed to detect symptoms of ASD in infants and toddlers aged 17 to 37 months. The *BISCUIT-Part 1* is comprised of 62 items that are scored on a 3-point Likert scale. The parent/caregiver of the child is instructed to rate each item in comparison to other same-aged children as: “0”– not different; no impairment, “1”– somewhat different; mild impairment, or “2”– very different, severe impairment. The total *BISCUIT-Part 1* score is calculated by adding each item score. A total score between 0-16 is categorized in the “No ASD/Atypical Development” range, a total score between 17-38 is categorized in the “Possible ASD” range, and a total score between 39-124 is categorized under “Probable ASD” range. Thus, children who receive a cut-off score of 17 or higher are considered at risk for ASD and should receive further assessment.

The *BISCUIT-Part 1* has been found to have strong psychometric properties, with an internal reliability of .97 (Matson et al., 2009). An exploratory factor analysis of the *BISCUIT-1* yielded three distinct factors: Socialization/Nonverbal Communication (S/NVC), Repetitive Behavior/Restricted Interests (RRBI), and Communication (Matson et al., 2010). Internal consistency of each factor was high, with a Cronbach's alpha of .93 in Factor 1, Cronbach's alpha of .90 in Factor 2, and Cronbach's alpha of .87 in Factor 3. The S/NVC subscale is comprised of 24 items, the RRBI subscale is comprised of 23 items (subscale items are displayed in Table 2), and the Communication subscale is comprised of 7 items, with item-total correlations ranging from .320-.702 (Matson et al., 2010).

Table 2. *BISCUIT-Part 1*, Repetitive Behavior/Restricted Interests subscale items

| <i>BISCUIT-Part 1</i> item number and description |
|---|
| 42. Abnormal fascination with the movement of spinning objects |
| 39. Interest in a highly restricted set of activities |
| 33. Sticking to odd routines or rituals that don't have purpose or make a difference |
| 58. Abnormal, repetitive motor movements involving entire body |
| 48. Becomes upset if there is a change in routine |
| 34. Abnormal preoccupation with parts of an object or objects |
| 55. Limited number of interests |
| 4. Engages in repetitive motor movements for no reason |
| 49. Needs reassurance, especially if events don't go as planned |
| 57. Abnormal, repetitive hand or arm movements |
| 27. Restricted interests and activities |
| 43. Curiosity with surroundings |
| 30. Reaction to sounds and sights |
| 6. Prefers food of a certain texture or smell |
| 11. Reactions to normal, everyday sounds |
| 41. Use of facial expressions |
| 38. Expects others to know their thoughts, experiences, and opinions without communicating them |
| 13. Reaction to normal, everyday lights |
| 61. Isolates self |
| 44. Saying words or phrases repetitively |
| 29. Eye-to-eye gaze |
| 8. Maintains eye contact |
| 26. Display a range of socially appropriate facial expressions |

The *Vineland Adaptive Behavior Scales, Third Edition (VABS-3)*; Sparrow, Cicchetti, & Saulnier, 2016) is an assessment tool designed to aid in the assessment of intellectual and developmental disabilities. There are three forms of the *VABS-3*, including the Interview Form, Parent/Caregiver Form, and Teacher Form. The Interview Form is administered by the examiner using a semi-structured interview method, and the examiner rates each item based on the parent/caregiver's responses. Items are rated on a 3-point Likert scale, according to the child's ability to complete a task. A rating of "0" indicates that the child is never able to perform the task, a "1" indicates that the child is sometimes able to perform the task, and a "2" indicates that the child is usually able to perform the task. Some items are rated as "yes" or "no". The items are scored to yield an overall adaptive behavior composite (ABC) score and four subdomain scores: Communication (COMM), Daily Living Skills (DLS), Socialization (SOC), Motor Skills (MOT). The ABC, COMM, DLS, SOC, and MOT scores will be used in the present study as a measure of a child's adaptive functioning.

The *Childhood Autism Rating Scale, Second (CARS2)*; Schopler, Van Bourgondien, Wellman, & Love, 2010) is an instrument used to assist in the diagnosis of ASD. The *CARS2* was designed to identify children 2 years and older with mild to severe symptoms of ASD. Additionally, the *CARS2* has been found to differentiate among children with ASD and children with other developmental disabilities. The *CARS2* measures functioning in 15 categories: Relating to People; Imitation; Emotional Response; Body Use; Object Use; Adaptation to Change; Visual Response; Listening Response; Taste, Smell, and Touch Response and Use; Fear or Nervousness; Verbal Communication; Nonverbal Communication; Activity Level; Level and Consistency of Intellectual Response; General Impressions.

The clinician rates the items based on direct observation, parent or caregiver report, and/or other sources of information (e.g., medical records, teacher reports). Each item is scored on 4-point scale: a score of “1” indicates no impairment/normal development, “2” indicates mildly abnormal behavior, “3” indicates moderately abnormal behavior, and “4” indicates severely abnormal behavior. The item scores are summed to produce a total score (i.e., severity rating), which can range from 15 to 60. The total score is then used to assign a severity group: a total score between 15 and 29 indicates “Minimal-to-No Symptoms of ASD”, a total score between 30 and 36 indicates “Mild-to-Moderate Symptoms of ASD”, and a total score of 37 and higher indicates “Severe Symptoms of ASD”. Reliability and validity evidence for the *CARS2* is unavailable; however, the original *CARS* (Schopler, Reichler, & Renner, 1986), has demonstrated high internal consistency (reliability coefficient alpha of .94) and validity ($r = .84$; Schopler, Reichler, DeVellis, & Daly, 1980).

Procedure

The Louisiana State University institutional review board and the Office for Citizens with Developmental Disabilities (OCDD) of the State of Louisiana approved the study prior to data collection. The *BISCUIT*, *Vineland-3*, and *CARS2* were administered by graduate student clinicians as part of the formal assessment of developmental functioning, which was comprised of a parent/caregiver interview, administration of standardized measures, and direct observation of the child. All evaluations were conducted by graduate student clinicians and supervised by a licensed clinical psychologist. Prior to the start of the assessment, informed consent to participate in research was obtained from the parent or caregiver of the child receiving the evaluation. Personal identifiers (e.g., name, date of birth) were removed from the database prior to analyses.

Statistical Analyses

Power analyses were conducted in G*Power (Faul, Erdfelder, Buchner, & Lang, 2009) to determine a sufficient sample size. Using an alpha of 0.05, a power of 0.80, and a medium effect size of 0.25 for a multiple regression with 6 predictor variables, the power analysis identified a sample size of 62 to be adequate.

All statistical analyses were performed using SPSS 26.0. Bivariate and multivariate analyses were conducted to answer the following research questions: (1) Which demographic factors and adaptive skill domains predict RRBI severity?, (2) Do parent-reported ratings of socialization, communication, and RRBI severity predict diagnostic ASD severity levels?, and (3) Which factors (i.e., *BISCUIT-Part 1* items, *CARS-2* total score and subscale scores, *VABS-3* composite score and subdomain scores) are significantly associated with ASD severity level?

A stepwise multiple regression was conducted to determine the predictive influence of several independent variables on RRBI severity. Predictor variables included MOT, SOC, COMM, and DLS subscale scores from the *VABS-3*, and the dependent variable was the *BISCUIT-Part 1* RRBI subscale score. A logistic regression was conducted to determine which *BISCUIT-Part 1* subscales (i.e., S/NVC, RRBI, Communication) predicted diagnostic ASD severity level. The *BISCUIT-Part 1* subscale scores were the independent (predictor) variables and ASD severity level was the dependent variable. Finally, a series of Spearman's rank-order correlations were conducted to assess the strength of the relationship between several factors (i.e., *BISCUIT-Part 1* items, *CARS2* total score, *CARS2* severity group, *CARS2* subscale scores, *VABS-3* ABC, *VABS-3* subdomain scores) and ASD severity level.

Results

RRBIs and Adaptive Functioning

To identify influential predictors for the stepwise multiple regression model, Spearman's rank correlations were first conducted to examine the strength of the association between the six potential predictor variables and the dependent variable. Four variables (i.e., MOT, SOC, COMM, and DLS of the *VABS-3*) had significant negative correlations with RRBI severity. Two variables (i.e., age, gender) were not significantly associated with RRBI severity. See Table 3 for Spearman's correlation coefficients. Therefore, MOT, SOC, COMM, AND DLS were included in the regression model and age and gender were excluded from the model.

Table 3. Correlations between RRBI severity and potential independent variables for the stepwise regression model

| Variables | ρ |
|--------------------|----------|
| Age | -.119 |
| Gender | -.055 |
| <i>VABS-3- MOT</i> | -.423*** |
| <i>VABS-3- SOC</i> | -.615*** |
| <i>VABS-3- COM</i> | -.307** |
| <i>VABS-3- DLS</i> | -.356*** |

Note: $N=89$. * $p \leq .05$, ** $p < .01$, *** $p \leq .001$

A stepwise multiple regression was run to determine if the addition of MOT, SOC, COMM, and DLS, as measured by the *VABS-3*, improved the prediction of RRBI severity scores, as measured by the *BISCUIT-Part 1* RRBI subscale score. See Table 4 for descriptive statistics for each *VABS-3* subdomain score and RRBI subscale score.

There was independence of observations, as assessed by a Durbin-Watson statistic of 2.194. According to Field (2013), the Durbin-Watson statistic can range from 0 and 4, with a value of approximately 2 indicating independence of residuals. There was linearity between the dependent variable and each of the independent variables, as assessed using a plot of studentized

residuals against the predicted values and partial regression plots. There was homoscedasticity, as assessed by visual inspection of a plot of studentized residuals versus unstandardized predicted values. Multicollinearity was assessed using the tolerance and variance inflation factors (VIF) values. There was no evidence of multicollinearity, as assessed by tolerance values of greater than .1 and VIF values of less than 10 (Hair, Black, Babin, & Anderson, 2014). There were no outliers, such that there were no studentized deleted residuals greater than ± 3 standard deviations, no leverage values greater than 0.2, and no values for Cook's distance above 1. The assumption of normality was met, as assessed by a Normal Q-Q Plot of the studentized residuals.

In Model 1, RRBI severity was significantly predicted by the SOC subdomain score alone, $R^2 = .40$, $F(1, 87) = 58.69$, $p = .00$. The addition of the MOT subdomain score (Model 2) also led to a statistically significant increase in variance, $\Delta R^2 = .005$, $F(2, 86) = 29.63$, $p = .00$. In Model 3, the addition of DLS also led to a statistically significant increase in variance, $\Delta R^2 = .008$, $F(3, 85) = 20.22$, $p = .00$. Finally, the addition of the COMM subdomain score (Model 4) led to a statistically significant change, $\Delta R^2 = .026$, $F(4, 84) = 16.68$, $p = .00$. See Table 5 for the stepwise multiple regression model prediction of RRBI severity using *VABS-3* subdomains.

Table 4. Descriptive statistics for stepwise regression model variables.

| Variables | M (SD) |
|--------------------|---------------|
| RRBI Total Score | 20.52 (9.11) |
| <i>VABS-3- MOT</i> | 82.64 (12.94) |
| <i>VABS-3- SOC</i> | 66.99 (13.64) |
| <i>VABS-3- COM</i> | 55.27 (20.38) |
| <i>VABS-3- DLS</i> | 70.81(15.40) |

ASD Severity Level

A logistic regression was conducted to determine if *BISCUIT-Part 1* subscale scores (i.e., S/NVC, RRBI, Communication) predicted clinician-assigned ASD severity levels. There was no

evidence of multicollinearity, as assessed by tolerance values of greater than .1 and VIF values of less than 10. The assumption of proportional odds was met, as assessed by a full likelihood ratio test comparing the fit of the proportional odds location model to a model with varying location parameters. Increased severity of RRBI did not significantly predict ASD severity level, $b = .06$, $\chi^2(1) = 3.47$, $p = .06$. Increased severity of SOC did not significantly predict ASD severity level, $b = -.03$, $\chi^2(1) = 1.336$, $p = .25$. Finally, increased severity of COMM did not significantly predict ASD severity level, $b = .08$, $\chi^2(1) = .58$, $p = .45$.

Table 5. Stepwise multiple regression for variables predicting RRBI severity

| Variable | RRBI Severity (RRBI Subscale Score) | | | | | | | | | | | |
|----------------|-------------------------------------|----------|--------------------|---------|----------|---------------------|---------|----------|---------------------|---------|----------|--------------------|
| | Model 1 | | | Model 2 | | | Model 3 | | | Model 4 | | |
| | R^2 | F | B (SE) | R^2 | F | B (SE) | R^2 | F | B (SE) | R^2 | F | B (SE) |
| | .40 | 58.69*** | | .41** | 29.63*** | | .42** | 20.22*** | | .44* | 16.68*** | |
| Constant | | | 48.92*** (3.78) | | | 51.80** * (5.04) | | | 51.24** * (5.06) | | | 53.88*** (5.15) |
| VABS-3 SOC | | | -.42*** (.06) | | | -.39*** (.07) | | | -.43*** (.07) | | | -.49*** (.08) |
| VABS-3 MOT | | | | | | -.06 (.07) | | | -.09 (.07) | | | -.08 (.07) |
| VABS-3 DLS | | | | | | | | | .07 (.07) | | | -.01 (.075) |
| VABS-3 COMM | | | | | | | | | | | | .11* (.06) |

Note: $N=89$. * $p \leq .05$, ** $p < .01$, *** $p \leq .001$

Finally, a series of Spearman's rank-order correlations were conducted to examine relationships between several variables and clinician-assigned ASD severity level. Given the large number of comparisons in this analysis, a Bonferroni adjustment for multiple comparisons was made, with an adjusted alpha level of $p \leq .001$. Table 6 displays the Spearman's correlation coefficients for each variable examined. On the *BISCUIT- Part 1*, no items or subscales were found to be significantly correlated with ASD severity level. Regarding the *CARS2*, the Body Use ($\rho(91) = .341$, $p = .001$) and General Impressions ($\rho(91) = .303$, $p = .00$) subscales were

significantly positively correlated with ASD severity. Additionally, the *CARS2* severity group ($\rho(91) = .359, p = .00$) was significantly positively correlated with ASD severity. On the *VABS-3*, *COMM* ($\rho(87) = -.328, p = .001$), *MOT* ($\rho(87) = -.336, p = .001$), and *ABC* ($\rho(87) = -.362, p = .00$) were significantly negatively correlated with ASD severity. No other variables were significantly correlated with ASD severity.

Table 6. Correlations between *BISCUIT-Part 1*, *CARS2*, and *VABS-3* variables and ASD severity

| Variable | Rho (ρ) | Variable | Rho (ρ) |
|------------------------|----------------|------------------------|----------------|
| <i>BISCUIT- Part 1</i> | | <i>BISCUIT- Part 1</i> | |
| Item 1 | -.071 | Item 46 | -.020 |
| Item 2 | -.038 | Item 47 | 0.110 |
| Item 3 | -.068 | Item 48 | -.040 |
| Item 4 | .072 | Item 49 | .071 |
| Item 5 | .000 | Item 50 | .179 |
| Item 6 | .206* | Item 51 | .089 |
| Item 7 | -.005 | Item 52 | -.020 |
| Item 8 | -.106 | Item 53 | .199 |
| Item 9 | .104 | Item 54 | .104 |
| Item 10 | -.030 | Item 55 | .032 |
| Item 11 | .073 | Item 56 | .057 |
| Item 12 | .102 | Item 57 | .234 |
| Item 13 | .117 | Item 58 | .175 |
| Item 14 | -.111 | Item 59 | .031 |
| Item 15 | -.060 | Item 60 | .129 |
| Item 16 | -.071 | Item 61 | .016 |
| Item 17 | -.094 | Item 62 | .005 |
| Item 18 | -.049 | S/NVC Score | .011 |
| Item 19 | -.071 | RRBI Score | .122 |
| Item 20 | -.098 | Communication Score | -.004 |
| Item 21 | .034 | Total Score | .070 |
| Item 22 | .149 | <i>CARS-2</i> | |
| Item 23 | -.040 | Relating to People | .077 |
| Item 24 | -.065 | Imitation | .306 |
| Item 25 | .114 | Emotional Response | .150 |

(table cont'd.)

| Variable | Rho (ρ) | Variable | Rho (ρ) |
|------------------------|----------------|--|----------------|
| <i>BISCUIT- Part 1</i> | | <i>BISCUIT- Part 1</i> | |
| Item 26 | .278 | Body Use | .341* |
| Item 27 | .012 | Object Use | .302 |
| Item 28 | -.037 | Adaptation to Change | -.040 |
| Item 29 | -.086 | Visual Response | .184 |
| Item 30 | .091 | Listening Response | -.019 |
| Item 31 | .059 | Taste, Smell, and Touch Response and Use | .255 |
| Item 32 | .121 | Fear or Nervousness | .208 |
| Item 33 | .035 | Verbal Communication | .236 |
| Item 34 | .039 | Nonverbal Communication | .235 |
| Item 35 | -.076 | Activity Level | .081 |
| Item 36 | .097 | Level and Consistency of Intellectual Response | -.049 |
| Item 37 | .074 | General Impressions | .393* |
| Item 38 | .083 | Total Score | .313 |
| Item 39 | -.050 | Severity Group | .359* |
| Item 40 | .012 | <hr/> <i>VABS-3</i> | |
| Item 41 | .109 | COMM | -.328* |
| Item 42 | .136 | DLS | -.280 |
| Item 43 | -.079 | SOC | -.008 |
| Item 44 | .078 | MOT | -.336* |
| Item 45 | .075 | ABC | -.362* |

Note: $N=93$. $*p \leq .001$

Discussion

The present study examined several factors associated with both RRBI severity and diagnostic severity level among young toddlers and children with ASD. Consistent with previous studies (Cuccaro et al., 2003; Matson, Kiely, & Bamburg, 1997), the current study found that lower adaptive functioning scores were significantly associated with higher RRBI severity. Of the four adaptive subdomains examined in this study, the Socialization subdomain had the strongest correlation with RRBI severity ($\rho = -.615$), followed by Motor Skills ($\rho = -.423$), Daily Living Skills ($\rho = -.356$), and Communication ($\rho = -.307$). This study is among the first to examine the predictive influence of adaptive functioning on RRBI severity. As expected, the addition of Socialization scores into the regression model led to a significant increase in variance for RRBI severity ($R^2 = .40$). The addition of Motor Skills scores to the model led to a significant increase in variance ($\Delta R^2 = .005$). Significant changes with the addition of the Daily Living Skills scores ($\Delta R^2 = .008$) and Communication scores ($\Delta R^2 = .026$) were also found. Overall, the full model, including all four adaptive subdomains, was found to significantly predict RRBI severity in young children with ASD, with 44% of the variance in RRBI severity explained by adaptive functioning skills. This suggests that toddlers with deficits across adaptive skills are at risk for increased severity of RRBI symptoms.

In order to address the gap in the literature regarding how *DSM-5* ASD severity levels are determined, parent- and clinician-rated measures of behavior were analyzed to identify which variables predict and are associated with ASD severity level. First, the three subscales of ASD symptoms according to the *BISCUIT-Part 1* (i.e., *S/NVC*, *RRBI*, *Communication*) did not significantly predict ASD severity level group membership (i.e., *ASD-Level 1*, *ASD-Level 2*, *ASD-Level 3*). This is not consistent with a previous study that reported significant correlations

between parent-report ratings of RRBI severity and ASD severity level (Mazurek et al., 2018). Given the young age of the study participants (i.e., 17-37 months), it may be that parents are not yet sensitive to and/or concerned about the social and communication delays that are associated with ASD. Additionally, parents may not perceive restricted, repetitive behaviors as impairing or atypical. Indeed, the existing literature indicates that parents typically report symptoms that are not characteristic of ASD as first concerns of their children's development (Kozlowski, Matson, Horovitz, Worley, & Neal, 2011; Matheis et al., 2016). Nevertheless, parents and caregivers have been found to reliably identify ASD symptoms in children as young as 12-18 months of age (Gray & Tonge, 2005; Reznick et al., 2007). Therefore, it may be that clinicians are determining ASD severity level according to the severity of the core symptoms of ASD as well as other factors.

To evaluate other factors associated with ASD severity level, a series of Spearman's correlations were conducted. First, the individual *BISCUIT-Part 1* items, subscale scores, and total score were examined. Of these variables, no statistically significant correlations were found. When clinician-rated measures of ASD symptoms were evaluated, two significant positive correlations between the *CARS2* subscale and ASD severity level were found: Body Use ($\rho = .341$) and General Impressions ($\rho = .393$). The *CARS2* Severity Group ($\rho = .359$) was also significantly positively correlated with ASD severity level. Of these significant relationships, the Body Use subscale can be categorized under the RRBI domain, and the General Impressions subscale and Severity Group can be categorized as overall measures of ASD severity. Regarding adaptive functioning, lower scores on the COMM, MOT, and ABC were significantly associated with higher level of clinician-assigned ASD severity; however, the DLS and SOC scores were not.

Although there is limited research on factors associated with ASD severity level, the present results from the series of correlations are consistent with what has been reported in the existing literature. Significant relationships between clinician-assigned ASD severity level and motor-related categories (i.e., Body Use of *CARS2*, MOT of *VABS-3*) suggest that motor RRBs and motor delays may be associated with ASD severity. Indeed, retrospective studies have found that children who were later diagnosed with ASD were reported to have early motor delays during infancy and toddlerhood (Ozonoff et al., 2008). Therefore, motor delays may be an early indicator of risk for ASD (Bhat, Galloway, & Landa, 2012). Further, previous studies have found strong relationships between presence of RRBI and ASD severity levels (Dworzynski et al., 2009; Kuenssberg & McKenzie, 2011; Matson et al., 2009). It may be that severity of motor RRBs and delayed motor functioning are significantly impairing and therefore, may warrant more support.

Social skills, as measured by the *BISCUIT-Part 1*, *CARS2*, and *VABS-3* were not significantly associated with ASD severity level, which is consistent with Mazurek et al. (2018)'s findings that there was no association between parent and clinician ratings of social communication and ASD severity rating. This finding is surprising given that impairment in socialization is a hallmark of the ASD phenotype. This discrepancy may be explained by methodological limitations. For instance, different studies may be using different measures and constructs of socialization, which likely explains the mixed findings. Though no studies, to date, have directly investigated the relationship between adaptive functioning and ASD severity level, the present findings support the idea that other areas of functioning are conflating ASD severity (Mazurek et al., 2018). Indeed, clinicians may be assigning a more severe level of ASD to

young children who have pervasive skill deficits, as those children require more substantial support and intervention.

The current study has several limitations that should be considered. First, two of the measures used in the study (i.e., *BISCUIT-Part 1*, *VABS-3*) relied on parent report. Though parents have been found to be reliable reporters of their children's behavior, there are several parental factors (e.g., cultural background, level of education, stress level, coping skills, social support) that may influence parents' perceptions of appropriate social, communication, and adaptive skills. Thus, researchers may consider investigating factors related to RRBI and ASD severity level while controlling for parent-specific factors. Second, RRBI severity was measured according to the *BISCUIT-Part 1* RRBI subscale, which includes various topographies of RRBI. A more fine-grained analysis of evaluating the factors associated with specific topographies of RRBI is warranted. Though no assumptions for statistical analyses were violated, there was an unequal distribution of participants when grouped by ASD severity level, with 6 participants diagnosed with ASD-Level 1, 24 participants diagnosed with ASD-Level 2, and 47 participants diagnosed with ASD-Level 3. Future studies should investigate factors associated with ASD severity level using more equal distributions of participants per severity group. In most cases, the graduate student clinician that administered and scored the *CARS2* assigned the ASD severity level rating, with the supervision of a licensed clinical psychologist. This may be a possible confound in the study. Therefore, future studies should have a clinician complete the clinician-rated measure and another clinician determine the diagnostic ASD severity level.

Despite these limitations, the present study fills a gap in the existing literature and contributes to the understanding of various factors that are associated with RRBI severity and ASD severity level. Taken together, the present findings have several implications for the early

assessment and treatment of RRBI and ASD-related symptoms. In regard to adaptive functioning and RRBI severity, young children with adaptive functioning deficits should be assessed for ASD at an early age and subsequently provided with targeted interventions designed to increase adaptive skills and reduce the severity of RRBI. This would ultimately mitigate the detrimental effects of RRBI on learning and daily functioning. Given the discrepancies between the present findings and the existing literature regarding the relationship between skill domains and ASD severity level, more research is needed in order to improve our understanding of the multitude of factors associated with the severity of ASD symptoms (e.g., intellectual functioning, communication skills, executive functioning skills, socioeconomic factors, cultural factors, gender, age). This would allow researchers and clinicians to identify specific areas in which a child needs support and determine the level(s) of support for specific skill domains. Additionally, RRBI and ASD symptom severity should be studied across development to evaluate the developmental trajectory of RRBI, changes in diagnostic ASD severity level, differential responses to treatment, and treatment outcomes in young children and adolescents with ASD.

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Vita

Esther Hong, born in Los Angeles, California, received her bachelor's degree from the University of Southern California. Following her undergraduate career, she worked as a behavior therapist for children and adolescents with autism spectrum disorder (ASD). She also worked as a research coordinator and conducted research on treatment outcomes in children and adolescents with ASD. In order to continue her clinical and research work, she decided to enter the Department of Psychology at Louisiana State University. Upon completion of her master's degree, she will begin work on her doctorate degree.