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Clinical Assessment of Repetitive and Restricted Behaviors in Toddlers on the Autism Spectrum

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Clinical Assessment of Repetitive and Restricted Behaviors in Toddlers on the Autism Spectrum

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Abstract

Repetitive and restricted behaviors (RRBs) are core symptoms of ASD and often emerge in the first year of life. It is important to understand how best to integrate diagnostically relevant information from multiple methods of assessment in clinical settings. The study aims to characterize the association between parent report and clinical observation of RRBs, compare the utility of each method for eliciting information on RRBs, and characterize the presentation of RRBs in toddlers at the time of initial diagnosis. Participants were 65 children ages 18-39 months diagnosed with ASD. Associations between a parent report rating scale, clinician-driven parent interview, and clinician observation were determined. Correlations were run for each of six behavioral subdomains. Rate of behavior endorsement on each measure was compared. Correlations were used to determine association between age and rate of RRBs.

In most behavioral subdomains, parent report of RRBs on a behavioral rating scale and clinical interview were consistent. Overall, parent report offers valuable information on RRBs that go beyond what is typically observed by clinicians in an evaluation. However, clinical observation appears to be particularly important for identifying sensory behaviors. Results indicate that both parent report and observation contribute to a more complete representation of RRBs and should be included in diagnostic assessment.

Clinical Assessment of Repetitive and Restricted Behaviors in Toddlers on the Autism Spectrum

Introduction

Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterized by deficits in social communication and the presence of atypical restricted and repetitive behaviors with an onset in early childhood. Restricted and repetitive patterns of behavior (RRBs) are core features of the clinical presentation of ASD (American Psychiatric Association, 2013). In the DSM-5, RRBs are characterized as repetitive behaviors, insistence on sameness, restricted interests, and hyper- and hypo-reactivity to sensory input. Repetitive behaviors may include stereotypic movements, such as hand-flapping or body-rocking, repetitive use of objects during play, or repetitive speech. Insistence on sameness is characterized by strict adherence to routines, inflexibility regarding change in routine, or ritualized behavior. Restricted interests are abnormally intense, fixated interests that may be highly distracting and challenging to redirect. Individuals with ASD may also exhibit hyper- or hyposensitivity to sensory stimuli in their environment, including lights, sounds, touch, temperature, or texture. The presence of these behaviors is essential for making a diagnosis of ASD, with at least two types of RRBs required in the current DSM-5 (American Psychiatric Association, 2013).

Subtypes of RRBs

While RRBs make up a single domain within diagnostic criteria, these behaviors are highly heterogeneous. In both clinical work and in research, RRBs are often conceptualized as including ‘lower order’ and ‘higher order’ repetitive behaviors (Turner, 1999). Higher order behaviors involve a cognitive component, while lower order behaviors refer to sensory motor symptoms and stereotypies (Richler, Bishop, Kleinke, & Lord, 2007). This structure is supported in the literature. Factor analyses of the Autism Diagnostic Interview, Revised (ADI-R) have

identified two factors of insistence on sameness (IS) and repetitive sensory motor behaviors (RSM) (Cuccaro et al., 2003; Szatmari et al., 2006). The IS domain includes resistance to change in one's routine or environment as well as compulsions and ritualized behavior. The RSM domain consists of stereotypic and repetitive movements, including body rocking and finger mannerisms, as well as repetitive use of objects and sensory interests. Within this model, circumscribed interests and preoccupations are not included (Szatmari et al., 2006). Factor analysis of the Repetitive Behavior Scale, Revised (RBS-R), a rating scale designed to capture a variety of RRBs in individuals with ASD, further divides RRBs into subgroups. Here, there are more items to describe behavior, and more distinct subtypes were identified. Hooker and colleagues (2019) found evidence for a five-factor model, which replicated similar results from Miranda et al. (2010), Lam and Aman (2007) and Bishop et al. (2013). Here, distinct factors include stereotypic behavior, self-injurious behavior, compulsive behavior, ritualistic and sameness behavior, and restricted interests. This is a shift from the previous two-factor model in that insistence on sameness behaviors, or 'higher order' RRBs, are further divided into the subgroups of compulsive behavior and ritualistic sameness behavior. Motor, object, and sensory stereotypies remain as one group, while self-injury and restricted interests appear as distinct subtypes (Hooker, Dow, Morgan, Schatschneider, & Wetherby, 2019).

Within the wider literature, sensory processing symptoms are often described as a distinct construct which can be further broken down into subtypes (K. K. Ausderau et al., 2016; Baranek et al., 2013; Ben-Sasson et al., 2009). While sensory symptoms have been recognized in association with ASD since the first descriptions of the diagnosis (Kanner 1943), these symptoms have only been included as core features in diagnostic criteria in the most recent DSM-5. Sensory processing refers to a person's ability to organize and interpret sensory

information (Germani et al., 2014). For individuals with ASD, atypical sensory responsiveness patterns have been described in four factors: hypo-responsiveness, hyper-responsiveness, sensory interests, repetitions, and sensory seeking behaviors, and enhanced perception (K. Ausderau et al., 2014). These four subtypes present across sensory modalities, including auditory, visual, tactile, olfactory, gustatory, and vestibular responses, as well as social and non-social contexts. Further, they frequently co-occur in individuals, contributing to great heterogeneity in symptom presentation (K. Ausderau et al., 2014). Several studies posit that some types of sensory symptoms may better distinguish ASD from other diagnostic groups, particularly hypo- and hyper-responsiveness (Baranek et al., 2013; Ben-Sasson et al., 2009; Joosten & Bundy, 2010; Rogers & Ozonoff, 2005). Thus, sensory symptoms represent yet another construct that contributes to the variable presentation of RRBs in individuals.

RRBs in Infancy and Toddlerhood

RRBs in infancy are not found exclusively in children with ASD or another neurodevelopmental disorder (Ozonoff et al., 2008; Ozonoff, Heung, Byrd, Hansen, & Hertz-Picciotto, 2008; Wetherby et al., 2004; Zwaigenbaum et al., 2005). Many of these behaviors are also observed in typical development, particularly repetitive object use and motor stereotypies. Researchers have hypothesized that these behaviors may be adaptive at an early age and promote cognitive development and problem solving in typically developing infants (Thelen, 1979; Thelen, 1981). Once the infant develops more adaptive skills to successfully explore their environment, however, RRBs decrease considerably and are replaced with more goal-directed behavior (Evans et al., 1997). Several studies, however, have documented significant differences in the number and severity of RRBs in children with ASD by the age of two years. Using parent report data from the ADI-R, Richler and colleagues (2007) found that repetitive sensory motor

behaviors (RSM) were highly prevalent in a sample of two-year-old children with ASD, while insistence on sameness (IS) was less common. Children in the ASD group had significantly more RSM than age-matched peers with other developmental delays or typical development. In this sample, children with ASD had an average of three RSM behaviors (Richler et al., 2007). Similarly, several studies have documented increased rates of RRBs in children 18 to 24-months old (Joseph, Thurm, Farmer, & Shumway, 2013; Kim & Lord, 2010; Richler, Huerta, Bishop, & Lord, 2010; Watt, Wetherby, Barber, & Morgan, 2008; Wetherby et al., 2004; Wolff et al., 2014; Wolff et al., 2019). In a sample of 18- to 24-month-old children, Watt et al. (2008) found higher frequency and longer duration of RRBs in the ASD group compared to developmentally delayed and typically developing controls. These behaviors included repetitive object use, such as banging, tapping, flipping, spinning, or wobbling objects, and repetitive body movements, such as stiffening and posturing with the hands and fingers, rubbing the body, and banging surfaces. Further, early RRBs predicted ASD symptom severity at age three years. Using the Autism Diagnostic Observation Schedule (ADOS), Kim and Lord (2010) found stable differences between ASD and other groups in all measured RRB subtypes, including sensory interests, hand and finger posturing, complex mannerisms, and repetitive behaviors. These differences in both the prevalence and severity of RRBs were present across all ages, from as young as eight-months to 56-months of age. Thus, RRBs are considered a reliable diagnostic feature in children at least two-years and older.

It was previously thought that repetitive behaviors were not widely present before this developmental age, as RRBs had not been identified via retrospective parent report data and home video analyses of high-risk infants (Cox et al., 1999; Lord, 1995; Werner & Dawson, 2005). A few studies, however, have considered the presence of specific repetitive behaviors as

early as the first year of life, and there is increasing data that suggest that RRBs occur prior to age two. Using direct observation methods and longitudinal data, Ozonoff et al. (2008) found that children who went on to receive a diagnosis of ASD exhibited significantly more repetitive object use at age 12 months compared to children with a developmental delay or typical development. These behaviors included increased spinning and rotating of objects and atypical visual exploration of objects. Unusual visual exploration was the most common symptom, seen in 77% of the sample (Ozonoff et al., 2008). This study is significantly limited by its small sample size of nine 12-month-old infants with ASD; however, it was among the first prospective studies on RRBs in ASD. In another study on ASD symptoms in the first year of life, Zwaigenbaum et al. (2005) reported that in addition to a variety of early social communication symptoms, sensory-oriented behaviors predicted diagnostic status at 24-months. More recently, Wolff and colleagues (Wolff et al., 2014; Wolff et al., 2019) found that RRBs across all subtypes were significantly elevated in parent report data as early as 12-months in children who later received a diagnosis of ASD compared to typically developing controls. The symptoms included stereotyped and ritualistic and restricted behavior (Wolff et al., 2014), as well as atypical sensory responsivity and sensory interests (Wolff et al., 2019). These data support the hypothesis that features of repetitive behaviors may be present very early in a child's development, at least for children at increased genetic risk for ASD.

Developmental Trajectory

Autism is a developmental disorder, and while diagnostic stability over time is high, symptom presentation changes based on the developmental level of the individual (Gotham, Pickles, & Lord, 2012). There are relatively few studies, however, on the developmental trajectory of RRBs. In general, the stability of RRBs depends on the type of behavior in question.

Lower order sensory motor behaviors have been observed to remain stable or even decrease over time in young children with ASD, while higher order insistence on sameness behaviors increase with age (Joseph et al., 2013; Kim & Lord, 2010; Richler et al., 2010). As a child grows and gains more developmental skills in both nonverbal and verbal domains, repetitive behaviors with more cognitive prerequisites appear. Kim and Lord (2010) found that RRB totals on the ADOS were similar for children less than one-year old up to five-years old, indicating stability over time. A few RRBs were found to increase with age; these included repetitive behavior (broadly defined to include insistence on sameness) and stereotyped language. In contrast, typically developing children's RRB scores decreased as children became older. This finding suggests a diverging developmental trajectory that becomes more pronounced over time. Similar results were found using parent report data on the RBS-R. A study by Wolff and colleagues (2014) concluded that RRBs appear to be stable as well as increasingly divergent from typically development between the ages of 12 and 24 months. Again, compulsive and ritualistic and sameness behaviors increased over time. Richler et al. (2010) observed children over a wider age range. In this study, children were assessed at age two, three, five, and nine. Overall, repetitive sensory motor behaviors decreased and became less severe with age, while insistence on sameness behaviors increased with age, further adding to the evidence that the developmental trajectories of RRBs are dependent on subtype.

The stability of RRBs over time appear to be somewhat individually variable and associated with cognitive abilities. Bishop et al. (2006) found that nonverbal IQ was associated with the presence of RRBs, and this relationship was stronger at older ages than younger ages in children younger than age 12. Further, repetitive sensory motor behaviors, including motor

stereotypies, are less likely to improve over time for individuals with lower nonverbal IQ and ASD (Esbensen, Seltzer, Lam, & Bodfish, 2009).

Understanding Function through Neural Underpinnings

While clinical presentations of RRBs are well-documented, neural underpinnings for these behaviors are much less understood. Recent studies describing neural morphology in connection with repetitive behaviors are important for better conceptualizing and understanding the function of these behaviors, which likely have implications for assessment and treatment. Within the domain of neural mechanisms, two primary, related areas of research have gained traction. These are differences in striatal growth and reward system circuitry activation in individuals with ASD. In regard to the former, several studies have documented an enlarged caudate nucleus in individuals with ASD. This difference in volume has been observed as early as two-years old with a widening gap in growth over time between children with ASD and typically developing children (Qiu et al., 2016). Faster striatal growth is correlated with increased higher-order RRBs in preschool age children (Langen et al., 2014). Langen et al. conclude that RBBs drive brain development in this region. Additionally, connective circuitry of the corpus collosum in infancy may be influential in the onset and development of repetitive behaviors. Increased corpus collosum area and thickness was reported in children with ASD beginning at six months of age, and these measurements were correlated with later repetitive behavior at age two years (Wolff et al., 2015).

The dorsal striatum and caudate nucleus are thought to be involved in learning and perception of reward (Kohls, Yerys, & Schultz, 2014). Restricted and repetitive behaviors and interests are considered to be influenced by altered, hyperactive reward circuitry responses to limited stimuli. Studies have demonstrated altered reward-system responsivity in children and

adults with ASD; individuals with ASD have shown diminished striatal response to social stimuli (Scott-Van Zeeland, Dapretto, Ghahremani, Poldrack, & Bookheimer, 2010) and monetary incentive, but not to stimuli relevant to circumscribed interests (Dichter et al., 2012; Kohls, Antezana, Mosner, Schultz, & Yerys, 2018). Thus, while RRBs in early childhood may influence the development of these brain regions, enhanced reward circuitry responses to these behaviors in turn reinforce and promote continued RRBs.

Interference with Social Development

It has long been thought that RRBs influence social outcomes for individuals with ASD. RRBs may be seen as strange by peers, making others hesitant to interact with affected individuals. Further, intense interests and difficulty exercising flexibility in activities and conversation may create a barrier to friendship seeking. Newer information about neural underpinnings and mechanisms may offer evidence that RRBs influence social outcomes even more directly and earlier in development by interfering with social learning.

One hypothesis by Kohls, Yerys, and Schultz (2014) suggests that higher order repetitive behaviors, such as insistence on sameness and restricted interests, develop in part because specific, favored stimuli are self-reinforcing by nature. Within their model, RRBs are rewarding because they satisfy a preference for predictability in the environment. In contrast, social situations are difficult to control and predict, and offer the individual more variable and less reliable reinforcements. Predictable reinforcement from non-social routine and interests leads to over-selection of preferred topics and, in turn, resistance to change. These reinforcement contingencies interfere with typical brain development, which may be reflected in the accelerated growth of the caudate in individuals with ASD. Within this model, the development of RRBs directly interferes with social development. RRBs take up resources that would otherwise go

towards social learning. Thus, RRBs are a central and dynamic feature of autism intrinsically related to social deficits.

Models of RRBs in ASD are not only crucial for better understanding the function and etiology of these behaviors, but also for conceptualizing and developing more effective interventions. For example, if the self-reinforcing nature of RRBs directly interferes with social learning over time and especially early in development, this hypothesis supports the use of intervention and redirection of these behaviors.

Assessment of RRBs

As our understanding of the presentation and course of RRBs grows, gaps in the literature remain regarding standardized assessment of RRBs (Harrop et al., 2014). Given the cascading influence of repetitive behaviors on social development, proper assessment is crucial not only for making a diagnosis, but also for informing treatment. Assessment of RRBs falls into two major categories: parent-reported information and clinician-observed behavior. Each of these methods for detection of RRBs have their relative strengths and weaknesses. Taken together, they may contribute to a more complete picture of repetitive behavior necessary to make a clinical diagnosis of ASD, though this hypothesis requires direct testing. Parent reported measures of RRBs include checklists and questionnaires, such as the Repetitive Behavior Scale, Revised (RBS-R) (Bodfish, Symons, Parker, & Lewis, 2000), and clinician administered interviews, including the Autism Diagnostic Interview and its revisions (ADI-R) (Lord, Rutter, & Le Couteur, 1994). Questionnaires like the RBS-R have been validated for use in toddlers (Lam & Aman, 2007; Mirenda et al., 2010) and provide an opportunity for dimensional assessment of RRBs (Wolff, Boyd, & Elison, 2016). Parent-report measures, however, are inherently limited by the parent's observation of the child's behavior and perception of this behavior as atypical.

Indeed, endorsement of concern for RRBs by parents prior to diagnosis varies widely. Despite apparent early onset of these behaviors, parents cite concerns about RRBs far less often than other concerns when presenting for a diagnostic evaluation (Herlihy, Knoch, Vibert, & Fein, 2015). The most commonly cited concerns that parents express about their child's development reference language and communication (De Giacomo & Fombonne, 1998; Richards, Mossey, & Robins, 2016). More specifically, delays in expressive language abilities appear to drive parental concerns of ASD (Karp, Ibanez, Warren, & Stone, 2017). Differences in parent endorsement of concern for RRBs may be influenced by previous experience, education, or having an older child with ASD (Herlihy et al., 2015). Further, parent-report questionnaires have limited utility in assessing severity of RRBs, especially in infancy and toddlerhood when it may be difficult for parents to distinguish atypical RRBs from behaviors expected in typical development. In addition, most measures query parents about a limited number of behaviors. Metrics of severity are not clearly defined, and ratings are subjective (Wolff et al., 2016). Clinician-administered interviews may mitigate these limitations, as they provide a trained clinician the opportunity to further prompt for suspected behavior and more reliably assess symptom severity; however, the most widely used clinician-administered diagnostic interview for ASD, the ADI-R, is not specifically designed for use in children under three years of age. Even with the use of developmentally appropriate diagnostic algorithms, assessment of RRBs is limited in scope (Cuccaro et al., 2003; Szatmari et al., 2006). Direct clinical observation represents the final method of detection of RRBs. A strength of this method is the direct observation and identification of atypical repetitive behavior by a trained clinician; however, the clinician's observation is inherently limited to behavior displayed during a limited time period. Even under conditions designed to elicit repetitive or restricted play behaviors, such as the Autism

Diagnostic Observation Schedule (ADOS) and its revision, it is likely that the clinician will not have the opportunity to observe all or most RRBs that the child uses, potentially making assessment of RRBs less comprehensive.

Due to inherent limitations in the various methods of assessment for RRBs, researchers and clinicians may use one or many of these methods to characterize a child's presentation. While a number of studies have identified incidence and developmental trajectory of RRBs in infancy and toddlerhood, no such studies have directly assessed or compared utility of different methods of assessment of RRBs for facilitating diagnosis. Given the low rates of reported concern for RRBs by parents and the importance of accurate, complete information for making a clinical diagnosis, it is important to understand which methods offer the highest yield of diagnostically relevant information in a clinical setting. This information may help clinicians to best integrate diagnostically relevant information from multiple methods of assessment. Further, changes in RRBs over time are defined in the literature as changes from lower order sensory motor behaviors to higher order RRBs with more cognitive requisites in later childhood (Richler et al., 2010). Less is known about trajectory of RRBs within the brief window of rapid developmental change from infancy to late toddlerhood.

Specific Aims

The current study seeks to (1) identify the association between multiple methods of assessment of RRBs, (2) determine which measures, if any, provide the highest rates of endorsement of RRBs by subdomain, and (3) characterize RRB presentation at the time of an initial diagnostic evaluation in children ages 18 to 40 months. With respect to the first aim, we expect the behavioral rating scale, clinical interview, and clinician observation not to be closely associated with one another. This may be due to differences in the environment and window for

observation, as well as potential differences in salience of these behaviors for parents and clinicians. Similarly, with respect to the second aim, given the low rates of reported concern for RRBs by parents, we expect clinician observation to yield the most information about a child's RRBs followed by clinician-driven interviews and finally parent-report questionnaires.

Finally, studies on the presentation of RRBs in toddlerhood show that repetitive sensory motor behaviors are typically stable over time, while higher order insistence on sameness behaviors increase with age (Kim & Lord, 2010; Wolff et al., 2014). In the present study, we use cross-sectional data to assess rates of RRBs over a relatively narrow developmental window of 18 to 40 months. We expect the rate of RRBs to be similar among children of all ages in all behavioral domains except higher order repetitive behaviors, which will be more prevalent in older children.

Methods

Participants

Participants were recruited as part of a larger study on the early detection of ASD. Children were screened using the Modified Checklist for Autism in Toddlers, Revised with Follow Up (M-CHAT-R/F) at their 18- or 24-month well-child visits at pediatricians' offices in Connecticut (Robins et al., 2014). When children screened positive on the M-CHAT-R/F, their parents were contacted by phone and a follow-up phone interview was conducted. Children who retained a positive screen after the follow-up interview were offered a no-cost developmental and diagnostic evaluation at the University of Connecticut with either a licensed clinical psychologist or behavioral developmental pediatrician. Families who were unable to travel to the clinic were offered an evaluation at their respective pediatrician's office. Of these children, 65 children were included in the present study. The current sample excludes children with missing data in primary

measures or any diagnosis other than ASD. An ASD diagnosis includes DSM-IV diagnoses of Autistic Disorder (AD), Autism Spectrum Disorder with low mental age (ASD Low MA), and pervasive developmental disorder, not otherwise specified (PDD-NOS).

Participants (n = 65) were 75% (n = 49) male (Table 1). Children ranged from 18.68 - 39.15 months of age at the time of evaluation. Average age at the time of evaluation was 24.03 months (SD = 4.40). 55 (84.61%) mothers and 10 (15.38%) fathers provided information for parent report measures. The sample was predominantly white (63.07%), and most mothers (76.92%) had some college or more education.

Procedure

Prior to clinical assessment, parents received measures in the mail including the Repetitive Behavior Scale, Revised (RBS-R) (Lam & Aman, 2007). Children were evaluated at the university clinic via direct assessment of current cognitive, language, motor and social functioning. Clinicians also interviewed parents to assess the child's adaptive functioning and ASD specific behaviors. Parents received immediate feedback regarding a diagnosis after all testing was completed and measures were scored. They received a written report that summarized testing and findings, diagnoses, and relevant recommendations in the mail approximately six to eight weeks after the evaluation.

The current study uses data collected from these evaluations. Measures that provide information about the child's RRB symptoms are used in the following analyses. Within each measure, individual items that captured the same behaviors were sorted into six behavioral subdomains: (1) sensory (2) hand and finger stereotypy (3) whole body stereotypy (4) combined stereotypy (5) self-injurious behavior (SIB) and (6) repetitive behavior. Appendix 1 describes the items included in each subscale for each measure.

Measures

Toddler Autism Symptom Inventory (TASI). The TASI is a semi-structured clinician administered parent interview. The TASI includes questions about a child's communication, social behavior, and RRBs. Questions are based on ICD-10 criteria (World Health Organization, 2004) for ASD, and items are scored to indicate either the presence of absence of symptoms. Preliminary data suggest good reliability and validity. Sensitivity (0.69) and specificity (0.89) for a clinical best estimate diagnosis of ASD in toddlers is high and comparable to other clinical interview measures (Coulter, 2019).

Autism Diagnostic Observation Schedule (ADOS). The ADOS (Lord et al., 2000) is a semi-structured, observational assessment of social interaction, play and imagination, communication abilities, and restricted and repetitive behaviors. The ADOS gathers information on an individual's social and communication abilities and other behaviors consistent with diagnosis of ASD. The ADOS consists of four modules selected as appropriate for the child's language level and chronological age. The majority of participants in the current sample (98.46%, n = 64) were tested using Module 1, designed for children without phrase speech. One child was tested with Module 2, designed for children with flexible phrase speech.

The ADOS includes scores in the domains of Reciprocal Social Interaction, Communication, Stereotyped Behaviors and Restricted Interests, and Play. Higher scores indicate greater symptom severity within each domain. Inter-rater reliability is considered to be high, with mean weighted kappas ($M\kappa_w$) of .78 in Module 1 and .70 in Module 2. Internal consistency is high in each module for the Social domain (α = .86 - .91), adequate in the Communication domain (α = .74 - .84), and adequate in the Stereotyped Behaviors and Restricted Interests domain (α = .63 - .65). Inter-rater agreement of classification (ASD verses

non-spectrum) is also high: 100% for Module 1 and 91% for Module 2. Sensitivity for classifying ASD verses non-ASD is .95-.97 for Modules 1 and 2; Specificity for classifying ASD verses non-ASD is .87-.94 for Modules 1 and 2 (Lord et al., 2000).

In the current study, the ADOS is used as a measure of direct clinician observation of the child's RRBs. The ADOS was administered at the time of the evaluation. Responses from only the Stereotyped Behaviors and Restricted Interests domain are used.

Repetitive Behavior Scale Revised (RBS-R). The RBS-R is a parent report behavioral rating scale designed to measure the presence and severity of a variety of repetitive and restricted behaviors and interests in children. The RBS-R consists of six sub-scales: stereotyped behavior, self-injurious behavior, compulsive behavior, ritualistic behavior, sameness behavior, and restricted behavior. Within each sub-scale, parents rate their child's behavior on a four-point likert scale (0 meaning the behavior does not occur, 1 meaning the behavior is a mild problem, 2 meaning the behavior is a moderate problem, and 3 meaning the behavior is a severe problem). In a sample of toddlers, internal consistency reliability was found to be acceptable for all sub-scales (total $\alpha = .92$; stereotyped $\alpha = .74$; self-injurious $\alpha = .75$; compulsive $\alpha = .79$; combined ritualistic and sameness $\alpha = .87$; restricted $\alpha = .79$) (Schertz, Odom, Baggett, & Sideris, 2016). In a different sample, inter-rater reliability was found to have intraclass correlation coefficients (ICC) ranging from .57 to .73 for all sub-scales. Further, items were correlated to their respective sub-scales, and correlations were moderate-to-high. The average correlation between items and the sub-scale total was 0.65 for ritualistic and sameness behavior, 0.57 for stereotypic behavior, 0.54 for compulsive behavior, and 0.61 for restricted interests (Lam & Aman, 2007).

Data Analytic Plan

Associations between measures were determined using Kendall's tau-b and point-biserial correlations where appropriate. Point-biserial correlations were selected for analyses including the TASI due to dichotomous outcome variables on this measure. Kendall's tau-b correlations were selected for determining association between the ADOS and the RBS-R, as both measures have ordinal scale data. The incidence of reported behavior was determined for each behavioral subdomain on each measure. This information was compared to determine which measures provided the highest yield of behavioral data. Relationship between RRB endorsement and age was determined using point-biserial correlations.

Data preparation. Item-level data were sorted into six respective behavioral subdomains (sensory behavior, hand and finger stereotypy, whole body stereotypy, combined stereotypy, self-injurious behavior (SIB) and repetitive behavior). Subdomains were selected to reflect the subcategories of behavior assessed in the measures. Here, the ADOS was the limiting measure, as the ADOS includes single scores for sensory behavior, stereotypies, SIB, and repetitive behavior. Of note, the ADOS does not include separate codes for hand and finger stereotypy and whole-body stereotypy, but rather combines these into a single subgroup. For the purposes of our comparisons, we assessed stereotypies both separately (on the TASI and RBS-R) and combined. Items were excluded if they assessed a specific behavior not captured on all other measures. When multiple items contributed to a single scale, Cronbach's alpha was used to determine acceptable reliability. Given acceptable reliability, a scale score was calculated. In the case of sensory behaviors, acceptable reliability for relevant items was not found; thus, items were recoded to reflect a single dichotomous score indicating either the presence or absence of sensory symptoms. That is, if responses indicated the presence of a sensory symptom, the child received

a score of 1 in this domain. These analyses were run on IBM SPSS Statistics, Version 26 (IBM Corporation, 2018).

Results

Aim 1: Association Between Measures

Scale reliability. Six subdomains of RRBs included two or more items. These items were combined to create subscales used for analysis. Of these, four subscales were created: SIB based on the RBS-R, repetitive behaviors based on the RBS-R, hand and finger stereotypies based on the TASI, and repetitive behaviors based on the TASI. Seven items on the RBS-R were selected for inclusion in the SIB subscale. Cronbach's alpha revealed acceptable internal consistency of this scale ($\alpha = .876$). The repetitive behavior scale based on the RBS-R was comprised of fifteen items with acceptable reliability ($\alpha = .826$). The hand and finger stereotypy scale based on the TASI is comprised three items with acceptable reliability ($\alpha = .748$). Nine items were included in the repetitive behavior scale based on the TASI with reliability approaching acceptability ($\alpha = .687$).

In the case of sensory behaviors, two items were selected for inclusion on both the RBS-R and the TASI to reflect the behaviors captured by the ADOS. For the sensory subdomain scores based on both the RBS-R and TASI, acceptable internal reliability was not established. Thus, items were collapsed into a single dichotomous variable reflecting either the presence or absence of reported atypical sensory behavior.

All other subscales were based on single items and did not require calculation of scaled scores.

ADOS and RBS-R. Correlations between measures are described in tables 1 through 7. Associations between the ADOS and RBS-R were determined using Kendall's tau-b correlations.

A moderate, statistically significant, positive correlation was found between the ADOS and the RBS-R in the domain of hand and finger stereotypy (τ_b = .31, p = .005). Correlations were non-significant in the domains of sensory, whole body stereotypy, combined hand and whole-body stereotypy, self-injury, and repetitive behaviors.

ADOS and TASI. Point-biserial correlation revealed a moderate, statistically significant correlation between the ADOS and the TASI in the domain of hand and finger stereotypy (r_{pb} = .30, p =.028). Correlations were non-significant in all other domains.

RBS-R and TASI. Point-biserial correlations were run to determine the association between subscales based on the RBS-R and the TASI. The two parent report measures were correlated with one another, with the exception of the items measuring sensory behaviors and combined stereotypies. RBS-R and TASI scores were correlated in the domains of hand and finger stereotypy (r_{pb}= .36, p = .004), whole body stereotypy (r_{pb}= .45, p< .001), self-injury (r_{pb}= .43, p< .001), and repetitive behavior (r_{pb}= .59, p< .001).

Aim 2: Compare Report of Behavior

The incidence of reported behaviors on the three measures is described in Table 8. The following results are reported in cases of nonsignificant correlations between measures, since these rates are understood to be meaningfully different. In the domain of repetitive behaviors, parents endorsed a higher incidence of repetitive behaviors on the TASI and especially on the RBSR-R compared to clinician observation on the ADOS. Clinicians observed repetitive behaviors in 75% of cases on the ADOS, while parents described repetitive behaviors in 94% of cases on the RBS-R and 77% of cases on the TASI. A similar pattern of reporting was found for self-injurious behaviors, with clinicians observing self-injurious behaviors in only 11% of cases and parents reporting SIB in 71% of cases on the RBS-R and 42% of cases on the TASI. In the

case of combined stereotypic movements, more stereotypic behaviors were reported on the RBS-R (71%), followed by the TASI (54%) and the ADOS (49%). Results were found in the opposite direction for sensory behaviors, with clinicians observing sensory-driven behaviors in 77% of cases compared to parent report in 65% of cases on the RBS-R and 48% of cases on the TASI.

Aim 3: RRBs by Age

Children were 18 to 39 months of age at the time of evaluation. Point-biserial correlations were run to determine the relationship between age and number of RRBs reported on the RBS-R, TASI, and ADOS. Correlations revealed no relationship between child's age and the number of RRBs in any of the behavioral subdomains on the TASI and ADOS. On the RBS-R, no relationship between age and RRBs was found for sensory behaviors, stereotypies, or self-injury. Point-biserial correlation revealed a weak but significant positive relationship between repetitive behaviors and age on the RBS-R (r_{pb}= .288, p< .05) such that repetitive behaviors were more likely to be observed in older children.

Discussion

The present study aimed to characterize the association between parent reported concern and clinical observation of RRBs to determine how to best integrate diagnostic information from multiple methods of assessment of RRBs in toddlers at the time of initial diagnosis.

Aim 1: Association Between Measures

We first sought to determine the level of association between multiple measures for the detection of RRBs in young children. This study compared a parent report behavioral rating scale, parent report clinical interview, and direct clinician observation. The RBS-R and TASI were significantly correlated on four out of six RRB subdomains. The method of obtaining parent report (behavioral rating scale versus clinician-driven interview) did not appear to impact

the information obtained for repetitive behaviors, self-injurious behaviors, hand and finger stereotypy and whole-body stereotypy. Thus, both the clinical interview and rating scale have comparable utility in obtaining information from parents about these behaviors and may be useful in a diagnostic evaluation. These measures were not correlated when hand and finger and whole-body stereotypies were combined as a single scale. This finding is surprising; in our sample, hand and finger stereotypies were correlated with whole body stereotypies on both the TASI and the RBS-R. Further, sensory behaviors were not associated on the RBS-R and the TASI. On both the RBS-R and the TASI, sensory items address sensory seeking behaviors and sensory sensitivity; however, hyper-sensitivity is described in more detail on the TASI, which may influence a parent's response. Neither method of parent report was closely aligned with clinician observation on the ADOS, except in the domain of hand and finger stereotypy. Hand and finger stereotypy may be more readily observable across contexts compared to other behaviors.

Aim 2: Compare Report of Behavior

The study's second aim was to determine the relative clinical utility of each measure in obtaining information about a child's RRBs. The incidence of reported behavior was determined for each measure and compared. When measures were not correlated by subdomain, the incidence of reported behavior was understood to be meaningfully different between the measures. Conversely, when measures were correlated by subdomain, report of these behaviors were understood to hang together and not reflect meaningful differences in report of behavior between the measures.

Contrary to the initial hypothesis, parent report on both the TASI and RBS-R revealed a higher number of RRBs in most domains compared to direct clinical observation on the ADOS.

Parent report identified RRBs beyond what was observed in the clinical evaluation in the domains of repetitive behavior, self-injurious behavior, whole body stereotypy, and combined hand and finger and whole-body stereotypy. Differences in rates of report may be explained by the limited opportunity for clinicians to observe these behaviors in a relatively brief window of time. Parents have the unique opportunity to observe their child's behavior often and over time, and this experience translates to valuable information for the diagnostic assessment. Further, it is possible that children were less likely to engage in certain repetitive behaviors, particularly repetitive and nonfunctional play, during the evaluation due to the presentation of new toys and stimuli. Children may play repetitively with specific, preferred toys at home, while sensory exploration and hand and finger stereotypies may be more common across multiple settings. Because of these concerns, the ADOS does not require repetitive behaviors to be coded in order to receive a score consistent with an autism spectrum disorder (Lord et al., 2000). These results further support this scoring consideration.

Previous studies have found that parents report concern for RRBs far less often than concerns for language and social behavior when presenting for diagnostic evaluation (Herlihy et al., 2015). At the time of initial evaluation, concerns about RRBs may be less salient for parents than other developmental concerns. The current results, however, indicate that when asked directly about these behaviors, either via behavioral rating scale or clinical interview, parents do offer substantial information on their observations of RRBs.

In contrast to other behavioral subdomains, sensory behaviors across measures were not correlated. Clinical observation on the ADOS revealed the most sensory behaviors, followed by the RBS-R and finally the TASI. Thus, clinical observation appears to be particularly important for identifying sensory behaviors. Sensory behaviors may be difficult for parents to identify or

discriminate as atypical, particularly in young children. For example, behaviors such as hand flapping or intense resistance to change may be more readily seen as atypical compared to rubbing surfaces or hyper-sensitivity to lights or texture. Further, parents may be less likely to report occasional sensory behavior if it is not interfering or salient in daily life.

Interestingly, only two items contributed to the sensory subdomain on the both RBS-R and the TASI, compared to fifteen items in repetitive behavior and eight items in SIB on the RBS-R. The TASI includes nine items on repetitive behavior. It is possible that parents benefit from more detailed questioning and specific examples in identifying certain behaviors or labeling those as atypical. This observation should be interpreted with caution, however, as this hypothesis was not directly tested in the current data and does not appear to be consistent across all subdomains.

Aim 3: RRBs by Age

Overall, rates of reported RRBs were not correlated with the age of the child, within the narrow age range sampled here. Repetitive behaviors, however, were correlated with age on the RBS-R only. On this measure, parents endorsed repetitive behavior for all children over the age of 20 months. In our analyses, the repetitive behavior domain includes rigid play, insistence on routine, and preoccupations, representing higher order RRBs. This finding is consistent with previous studies, which found that higher order RRBs, such as compulsive and ritualistic behavior, increase with age in early childhood, even between the first and second birthday (Richler et al., 2010; Wolff et al., 2014). Repetitive behaviors were not correlated with age on either the TASI or the ADOS, indicating an effect of measurement tool.

We found no effect of age for sensory behavior, motor stereotypies or SIB. These behaviors are thought to require less cognitive demand and maturity, and thus may be less likely to increase between 18 to 40 months.

Clinical Implications

Overall, these results suggest that parent report and clinical observation together contribute to a more complete characterization of a child's repetitive behavior symptoms. Results can guide clinical decision making on how best to integrate symptom information through multiple methods of assessment. As for obtaining parent report, results suggest that both the RBS-R and the TASI offer comparable information on RRBs, except in the domain of sensory behaviors where the rating scale slightly outperforms the interview. Sensory behaviors are best detected, however, by clinician observation on the ADOS. Given this discrepancy in identification, parents may benefit from increased education about sensory play and interests in order to better identify these behaviors in their children. It might also be helpful to develop more refined and detailed measures of sensory issues in young children or to add those to existing measures. The diagnostic evaluation provides a unique opportunity for clinicians to point out observed sensory behaviors to parents and offer recommendations on how to redirect sensory driven play. Better identification of RRBs leads to more individually tailored and effective treatment plans. Further, when categorized by these broad subdomains, type and frequency of RRB does not appear to change within this narrow age range. These findings underscore the importance of assessing for RRBs in each behavioral subdomain, even in very young children.

Limitations and Future Directions

The current study compares report of RRBs on the subdomain level but lacks comparison of specific behaviors on the item level. The ADOS only reports data on broad categories of

behavior, which limits this study's potential for more fine-grained comparisons across measures. It is possible that item-level analysis would reveal relevant differences in the identification of specific behaviors. For example, parents may be better able to discriminate sensory seeking behaviors than over- or under-reactivity to sensory stimuli or vice versa. Future research should include specific, item-level data that is comparable across data collection modality (e.g., rating scale, interview, observation) to allow for more detailed analysis, both in comparing methods of assessment and considering presentation by age.

Additionally, all parents in the current study agreed to attend a diagnostic evaluation and often had preexisting concerns about their child's development. Parents without concerns may be less likely to notice RRBs as atypical and respond affirmatively on diagnostic measures. For parents without expressed concern, clinician observation or more detailed measures of RRBs and sensory behaviors may play a more important role in identification of behaviors consistent with a diagnosis of ASD. Future work would benefit from assessment of preexisting parent concern and its impact on identification of behaviors.

Parent reported symptoms may be influenced by a parent's culture, education, or general understanding of ASD, as developmental expectations and interpretations of behavior may vary across cultural and ethnic groups. For example, one study documented that Puerto Rican mothers expected their children to reach certain social milestones, such as social smiling, later than white mothers (Pachter & Dworkin, 1997). In some Asian cultures, pointing with an index finger is less common and may not considered important in a child's development as in majority North American cultures (Zhang, J., Wheeler, J. J., & Richey, D., 2006). These differences have important implications for ASD detection, and there may be similar culturally dependent expectations for repetitive behavior. In this sample, the majority of parents participating in the

evaluation are college-educated, White mothers. Generalizability of these findings may be limited for diverse populations.

Further, our sample considers RRBs at the time of initial diagnosis using cross-sectional data. While this allows us to assess RRBs by age among children who have not yet received a diagnosis or intervention, it does not allow for consideration of individual developmental trajectory. In longitudinal samples, particularly once children have gained access to intervention, RRBs may change over time.

Conclusions

Both parent report and clinician assessment of RRBs are critical for complete, accurate diagnostic information. Between the behavioral rating scale and the clinical interview, the method of parent report did not influence data obtained. Parent repot on the RBS-R and TASI were consistent in in most behavioral subdomains. Clinical observation on the ADOS revealed fewer RRBs than parent report in repetitive behavior, self-injurious behavior, whole-body stereotypy, and combined stereotypy and comparable report on hand and finger stereotypy. Differences may be due to the brief observation window or the introduction of novel toys and a new environment in the evaluation. Notably, none of the measures were correlated for sensory behaviors. In this domain, clinician observation on the ADOS outperformed parent report on both the TASI and RBS-R. Overall, parent report offers valuable information on RRBs that go beyond what is typically observed by clinicians in an evaluation. However, clinical observation appears to be particularly important for identifying sensory behaviors. Results indicate that both parent report and observation contribute to a more complete representation of RRBs and should be included in diagnostic assessment.

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Table 1.

Participant Demographic Characteristics

Variable	Participants (n = 65)
Sex N (%)	
Male	49 (75.38%)
Female	16 (24.62%)
Age at Evaluation: M (SD)	24.03 (4.40) months
Range	18.68 - 39.15 months
Race or Ethnicity N (%)	
White or European American	41 (63.07%)
Black or African American	6 (9.23%)
Asian or Pacific Islander	5 (7.69%)
Hispanic or Latino	7 (10.77%)
Biracial	5 (7.69%)
Missing	1 (1.54%)
Maternal Education N (%)	
No high school degree/GED	6 (9.23%)
High school degree/GED	8 (12.31%)
Some college	27 (41.5%)
Bachelor's degree	15 (23.08%)
Advanced degree	8 (12.31%)
Missing	1 (1.54%)
Parent Relationship N (%)	

Mother	55 (84.61%)
Father	10 (15.38%)
Diagnosis N (%)	
Autistic Disorder	34 (52.31%)
ASD Low MA	3 (4.62%)
PDD-NOS	28 (43.08%)

Table 2.

Sensory Behavior Correlations

Variable	1	2	3
1. ADOS Sensory	-		
2. RBS-R Sensory	.22	-	
3. TASI Sensory	-.11	.24	-
4. Age	.06	.17	.23

Note: Total $N = 65$; *p < .05; **p < .01; ADOS= Autism Diagnostic Observation Schedule; RBS-R= Repetitive Behavior Scale, Revised; TASI= Toddler Autism Symptom Inventory

Table 3.

Stereotypy Hand Movements Correlations

Variable	1	2
1. ADOS Stereotypy	-	
2. RBS-R Stereotypy Hand	.31**	-
3. TASI Stereotypy Hand	.30*	.36**

Note: Total $N = 65$; *p < .05; **p < .01

Table 4.

Stereotypy Whole Body Movements Correlations

Variable	1	2
1. ADOS Stereotypy	-	
2. RBS-R Stereotypy Whole Body	.01	-
3. TASI Stereotypy Whole Body	.06	.45**

Note: Total $N = 65$; *p < .05; **p < .01

Table 5.

Stereotypy Combined Correlations

Variable	1	2	3
1. ADOS Stereotypy	-		
2. RBS-R Stereotypy Combined	.21	-	
3. TASI Stereotypy Combined	.24	.04	-
4. Age	.07	-.001	.09

Note: Total $N = 65$; *p < .05; **p < .01

Table 6.

Self-Injurious Behavior Correlations

Variable	1	2	3
1. ADOS Self -Injurious Behavior	-		
2. RBS-R Self-Injurious Behavior	.21	-	
3. TASI Self-Injurious Behavior	.24	.43**	-
4. Age	-.14	.21	-.02

Note: Total $N = 65$; *p < .05; **p < .01

Table 7.

Repetitive Behavior Correlations

Variable	1	2	3
1. ADOS Repetitive Behavior	-		
2. RBS-R Repetitive Behavior	.13	-	
3. TASI Repetitive Behavior	.05	.59**	-
4. Age	-.06	.29*	.08

Note: Total $N = 65$; *p < .05; **p < .01

Table 8.

Percent Behavior Endorsement by Measure

Behavior	ADOS	RBS-R	TASI
Sensory	77%	65%	48%
Stereotypy - Hand	N/A	65%	37%
Stereotypy – Whole Body	N/A	35%	38%
Stereotypy - Combined	49%	71%	54%
Self-Injurious Behavior	11%	71%	42%
Repetitive Behavior	75%	94%	77%

Note: Total $N = 65$

Appendix 1. Scales

Sensory Items

RBS-R	6: Sensory (Covers eyes, looks closely or gazes at hands or objects, covers ears, smells or sniffs items, rubs surfaces)
	22: Touch/ Tap; Need to touch, tap, or rub items, surfaces, or people
TASI	DSMV1: Does your child show unusual sensitivity to certain noises, textures, smells, tastes, or being touched, so that he/she is distressed by them? This might be apparent in refusing to wear certain clothes, eat foods of a certain texture or temperature, go certain places, cover his ears, etc.
	DSMV3: Does your child appear to seek out particular sensory experiences such as noises, textures, smells, tastes, or deep pressure, etc?
ADOS	1: Unusual sensory interest in play material/ person.

Hand and Finger Stereotypies

RBS-R	3: Hand/Finger (Flaps hands, wiggles or flicks fingers, claps hands, waves or shakes hand or arm)
TASI	R3A: Does your child flap her/his hands and/or arms?
	R3B: Does your child move her/his fingers or hands near her/his face or eyes in an apparently nonfunctional way?
	R3C: Does your child make other unusual or repetitive movements with her/ his hands or fingers?
ADOS	2: Hand and Finger or other complex mannerisms.

Whole Body Stereotypies

RBS-R	1: Whole Body (Body rocking, Body swaying)
TASI	R3E: Does your child engage in unusual movements with her/ his body (e.g., jumping repeatedly, spinning, pacing, crashing into furniture, bouncing from foot to foot, engaging in a "happy dance," tensing whole body, etc.)? When do these movements occur? Can your child be easily redirected?
ADOS	2: Hand and Finger or other complex mannerisms.

Self-Injurious Behavior

RBS-R	7: Hits self with body part (Hits or slaps head, face, or other body area)
	8: Hits self against surface or object (Hits or bangs head or other body part on table, floor, or other surface)
	9: Hits self with object (Hits or bangs head or other body area with objects)
	10: Bites Self (Bites hand, wrist, arm, lips, or tongue)
	11: Pulls (Pulls hair or skin)
	12: Rubs or scratches self (Rubs or scratches marks on arms, leg, face, or torso) 13: Inserts finger or object (eye-poking, ear-poking)

	14: Skin picking (Picks at skin on face, hands, arms, legs or torso)
TASI	DSMV4: Does your child engage in self-injurious behavior? (e.g., biting his/her hand, banging his/ her head)
ADOS	3: Self-Injurious Behavior

Repetitive Behavior

RBS-R	15: Arranging/ Ordering; arranges certain objects in a particular pattern or place; Need for things to be even or symmetrical
	27: Play/Leisure; Insists on certain play activities; follows a rigid routine during play; insists that certain items be present/available during play/leisure; Insists that other persons do certain things during play
	28: Communication/ Social interactions; Repeats same topics during social interactions: Repetitive questioning; Insists on certain topics of conversation; Insists that others say certain things or respond in certain ways during interactions
	29: Insists that things remain in the same place(s) (e.g., toys, supplies, furniture, pictures, etc.)
	31: Becomes upset if interrupted in what he/she is doing
	32: Insists on walking in a particular pattern (e.g., straight line)
	33: Insists on sitting at the same place
	34: Dislikes change in appearance or behavior of the people around him/her
	35: Insists on using a particular door
	37: Resists changing activities; difficulty with transitions
	38: Insists on same routine, household, school or work schedule everyday
	39: Insists that specific things take place at specific times
	41: Strongly attached to one specific object
	42: Preoccupation with part(s) of object rather than the whole object (e.g., buttons on clothes, wheels on toy cars)
	43: Fascination, preoccupation with movement/ things that move (e.g., fans, clocks)
TASI	R1A: Does your child have any interests that are unusual for a child her/his age (e.g., fans, toilets, hubcaps, lights, spinning objects)?
	R1C: Does your child play with her/his toys in a specific, routinized manner (e.g., lining up her/his dolls, putting Legos together only in a specific pattern)?
	R1D: Does your child like to carry around unusual items (e.g., needs to have something in both hands, carries spoons all day, etc.)?
	R2A: Does your child insist that routines be conducted in the same order (e.g., insists that a parent says a phrase in the same way every time, becomes distressed if parent takes alternate route home in the car, distressed by change in daycare routine)?
	R2B: If your child has rituals/ routines, does your child attempt to impose her/his routines or rituals on others (e.g., moving a sibling's toy

	cars so that they are in a specific pattern, requiring all who come into your house to remove hats)?
	R2C: Is your child distressed if minor changes occur in her/his environment that don't directly affect him/her (e.g., changing the color of your living room, cutting your hair, a parent getting new glasses)?
	R2D: Does your child become distressed or upset if there are minor changes in her/his immediate environment (e.g., getting new sheets for her/his bed, changing from shorts to long pants in autumn)?
	R4A: Does your child play with parts of toys in nonfunctional ways (e.g., spinning wheels of toy trucks instead of wheeling them on a surface, playing with the eyes or ponytail on a doll but not playing with the doll itself?)
	R4D: Does your child manipulate objects in nonfunctional ways (e.g., dangle shoelaces while looking at them, tapping toy cars on tables, flipping pages of books)? Can your child be easily redirected?
ADOS	4: Unusually Repetitive Interests or stereotyped behaviors.