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A Distinct Three-Factor Structure of Restricted and Repetitive Behaviors in an Epidemiologically Sound Sample of Preschool-Age Children with Autism Spectrum Disorder

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Abstract

Prior studies investigating restricted and repetitive behavior (RRB) subtypes within autism spectrum disorder (ASD) have found varied factor structures for symptom groupings, in part, due to variation in symptom measurement and broad sample age ranges. This study examined RRBs among 827 preschool-age children, ages 35 to 71 months, through an exploratory factor

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analysis of RRB items from the Autism Diagnostic Interview-Revised (ADI-R) collected through the Study to Explore Early Development. The factor structures of RRBs among children with confirmed ASD versus those with non-autism developmental concerns were qualitatively compared. Correlations between RRB factors and participant characteristics were examined in the ASD group. Three conceptually well-defined factors characterized as repetitive sensorimotor behaviors (RSMB), insistence on sameness (IS), and a novel stereotyped speech (SPEECH) factor emerged for the ASD group only. Distinct factors were supported by different clinical correlates. Findings have implications for improving differential diagnosis and understanding of ASD symptomatology in this age range.

Keywords

Autism spectrum disorder (ASD); Exploratory factor analysis; Preschool children; Restricted and repetitive behaviors (RRB)

Restricted and repetitive behaviors (RRBs) are a heterogeneous group of symptoms comprising a core feature of autism spectrum disorder (ASD). Previous studies have distinguished RRBs within ASD from those observed in typical development or in other psychiatric and neurodevelopmental disorders given the higher frequency, variety, and severity of RRBs observed among individuals with ASD (Barrett et al. 2004; Flores et al. 2011; Matson et al. 2009; Watt et al. 2008; Wiggins et al. 2009). RRBs in children with ASD often emerge by 24 months of age and become more apparent as atypical at 3 to 5 years of age (Elison et al., 2014; Ozonoff et al. 2008; Wolff et al. 2014) with specific RRBs varying in prevalence by age range. More precise understanding of RRB presentation in early childhood is of critical importance for early and accurate diagnosis of ASD and subsequent referrals to early intervention to optimize developmental outcomes.

The *Diagnostic and Statistical Manual of Mental Disorders* (DSM-5; American Psychiatric Association 2013) places greater emphasis on RRBs relative to earlier diagnostic schemes, describing four categories of RRB patterns within ASD: (1) stereotyped or repetitive motor mannerisms, use of objects, or speech; (2) rigid behaviors such as insistence on sameness, inflexible adherence to routines, or ritualized patterns of behavior; (3) circumscribed interests that are highly restricted, fixated, or unusual in focus; and (4) abnormal sensory responses including hyper- or hyporeactivity to sensory input. Inclusion of repetitive speech patterns and sensory processing abnormalities as core RRB symptoms within autism is new to DSM-5, and prior studies exploring the factor structure among young children have not always included the full range of these behaviors.

Factor analyses have commonly been employed to better understand RRB symptom presentation among individuals with autism spectrum diagnoses. Most studies have relied on caregiver-report of behaviors from the Autism Diagnostic Interview-Revised (ADI-R; Rutter et al. 2003), the Repetitive Behavior Scale, Revised (RBS-R; Bodfish et al. 2000), and the Repetitive Behaviour Questionnaire-2 (RBQ-2; Leekam et al. 2007). Many studies spanning a wide age range and using various measurement tools have found at least two distinct factors, usually characterized as repetitive sensory motor behaviors and insistence on sameness (Bishop et al. 2006; Cuccaro et al. 2003; Georgiades et al. 2010; Lidstone

et al. 2014; Szatmari et al. 2006; Richler et al. 2010). Yet, others have found three or more factors, including RRB factor categories such as repetitive behaviors, preoccupations, unusual sensory interests, insistence on sameness, compulsions, self-injury, and/or restricted interests (Bourreau et al. 2009; Hooker et al. 2019; Lam and Aman 2007; Lam et al. 2008). Findings have varied based on the measurement tools used; for example, Bishop et al. (2013) found two distinct factors for ADI-R items and five factors for RBS-R items.

Few studies have focused exclusively on young children using the ADI-R, which is considered to be a gold-standard diagnostic instrument for ASD. Richler et al. (2007) and Mooney et al. (2009) each found two factors consisting of repetitive sensory motor behaviors and insistence on sameness when examining RRB endorsements on 12 items from the ADI-R among samples including children with autism spectrum/pervasive developmental disorder diagnoses under three years of age and children aged 20–55 months, respectively. Richler et al. (2007) conducted a confirmatory factor analysis of ADI-R RRB items, excluding the circumscribed interest item due to the young age range of their sample (as this item is not administered to children under 3 years of age) and the verbal rituals item which was only administered to children with phrase speech. They added self-injurious behavior given that this behavior can be repetitive in nature, although it is not included in the ADI-R RRB section. The exploratory factor analysis conducted by Mooney et al. (2009) included ADI-R RRB items with the addition of self-injurious behavior and excluding items that were incompatible with the young age and language abilities of their sample. In a principal component analysis of 12 ADI-R items (similar to those analyzed in Richler et al. 2007) among children aged 24–48 months, Honey et al. (2008) found three emerging components including sensory motor behaviors, resistance to change, and unusual interests.

Studies using other tools to measure RRBs among young children with ASD include Leekam et al. (2007) who found four factors using the RBQ-2 (i.e., unusual sensory interests, repetitive motor movements, rigidity/adherence to routine, preoccupations with restricted patterns of interest) and Mirenda et al. (2010) who found that a three factor (i.e., stereotypy restricted, self-injurious, compulsive ritualistic sameness) or five factor (i.e., stereotypy, self-injurious, compulsive, ritualistic sameness, restricted) model provided a good fit using the RBS-R. Across factor analysis studies, the varying numbers of latent RRB factors may reflect the inclusion of specific RRB items and the age ranges of the samples (Leekam et al. 2011), and many were limited by a small sample sizes relative to the number of items examined.

Given prior research has demonstrated variation in the underlying factor structure of RRBs, that behavioral symptom presentation may vary as a function of age, and few studies of younger ages have examined the range of behaviors represented in updated ASD criteria, the present study aimed to further explore RRB characteristics specifically exhibited by preschool-age children with ASD through an exploratory factor analysis of ADI-R items that reflect the full range of RRBs described in DSM-5. The inclusion of children who were specifically preschool-aged was deemed to be important given that this is the age range in which a fuller range of RRBs become more apparent. Further, since RRBs are not exclusive to ASD, we compared RRB items and factor structures across age-matched groups with and without ASD to determine which behavioral features are more commonly reported

for those with ASD versus those with non-ASD developmental delays. While previous studies in this area have used relatively small convenience samples, the present analysis examined the factor structure of RRBs among a large, epidemiologically sound population of preschool-aged children with confirmed ASD features (ASD group) and children with nonspecific developmental concerns who do not meet ASD criteria (ASD-negative group).

Methods

Data Selection Procedures

Data were collected through the multisite epidemiologic Study to Explore Early Development (SEED; Schendel et al. 2012). Eligible children were born between September 2003 and August 2006, and resided in one of six study catchment areas (California, Colorado, Georgia, Maryland, North Carolina, and Pennsylvania). Children enrolled in SEED comprised three groups: those with confirmed ASD characteristics, those with developmental concerns or disorders other than ASD who were recruited from educational and health agencies, and those who are typically developing who were recruited from birth records. Research approval was obtained from the IRB at each site and caregivers provided written consent.

Enrolled children were screened for ASD with the Social Communication Questionnaire (SCQ; Rutter et al. 2003) using a lower threshold (scores ≥ 11) to maximize sensitivity (Wiggins et al. 2009). Children who had a prior diagnosis of ASD and/or scored ≥ 11 on the SCQ were asked to complete a comprehensive ASD evaluation. All others completed a limited developmental evaluation. If ASD behaviors were witnessed during the limited evaluation, the clinician could refer the child for the comprehensive evaluation. The present analysis is limited to those who completed a comprehensive ASD evaluation.

For the comprehensive evaluation, the Mullen Scales of Early Learning (Mullen 1995) was used to assess children's developmental levels. Information on behavioral characteristics was obtained through parent report using the Child Behavior Checklist (CBCL; Achenbach and Rescorla 2000) to assess internalizing and externalizing behaviors and presence of anxiety, mood, and ADHD concerns, and the Vineland Adaptive Behavior Scales, Second Edition (Vineland-II; Sparrow et al. 2005) to assess adaptive functioning. Research reliable clinicians administered gold-standard tools for assessing ASD (Gray et al. 2008) including the Autism Diagnostic Observation Schedule (ADOS; Lord et al. 1999), a semi-structured observational assessment, and the ADI-R, a standardized diagnostic interview with parents/caregivers to assess the presence and severity of autism symptoms. Information provided by caregivers on the ADI-R was coded by examiners, with lower ratings on the ADI-R indicating fewer concerns/less frequent and interfering atypical behaviors and higher ratings indicating more significant concerns/more frequent and interfering atypical behaviors. "Current" and "ever" ratings are specified on the ADI-R to capture behaviors that have recently been observed versus behaviors that may have been present previously in development. For this analysis, current ratings that were age- or language-level appropriate were examined.

For this analysis, children in the ASD group were those who completed the comprehensive ASD evaluation and met the SEED case definition (Wiggins et al. 2015) for ASD by meeting criteria on *both* the ADOS and ADI-R. Children who completed a comprehensive evaluation but did not meet final SEED classification for ASD (i.e. scored below the autism diagnostic cutoff on *both* the ADOS and ADI-R) were included as a clinically relevant comparison group of children with non-ASD developmental concerns (ASD-negative group). Figure 1 details how SEED subsamples were determined.

Participants

Participants for this study included 827 eligible children, 707 categorized as ASD and 120 classified as ASD-negative. Demographic and developmental characteristics for both samples are in Table 1. Participants ranged in age from 35–71 months and both groups had a similar mean age. Significant differences in demographics were found between groups, with the ASD group including higher percentages of males ($p < .001$), White/Non-Hispanic maternal race and ethnicity ($p < .001/p < .01$), and higher maternal education ($p < .001$). In terms of clinical characteristics, the ASD group showed greater levels of ADHD symptoms, externalizing problems, and internalizing problems reported on the CBCL as well as lower scores across developmental and adaptive measures (all $p < .001$).

RRB Item Selection

The present analysis examined a total of 13 ADI-R items to capture the full range of RRB items described in DSM-5, which included: (67) Unusual Preoccupations; (68) Circumscribed Interests (for subjects 3 years or older); (69) Repetitive Use of Objects or Interests in Parts of Objects; (70) Compulsions/Rituals; (71) Unusual Sensory Interests; (73) Abnormal, Idiosyncratic, Negative Response to Specific Sensory Stimuli; (74) Difficulties with Minor Changes in Subjects' Own Routines or Personal Environment; (75) Resistance to Trivial Changes in the Environment; (76) Unusual Attachment to Objects; (77) Hand and Finger Mannerisms; and (78) Other Complex Mannerisms or Stereotyped Body Movements. In addition, the following items were also added to reflect repetitive speech patterns now included as RRB features in DSM-5 ASD diagnostic criteria: (33) Stereotyped Utterances and Delayed Echolalia; and (39) Verbal Rituals. RRB items including Undue General Sensitivity to Noise (item 72) and Midline Hand Movements (item 79) were removed given their redundancy with other items in the RRB section (items 73 and 77, respectively). Circumscribed interests and verbal ritual items were retained in this sample of preschool-age children who were generally between the ages of 3–5 years.

Statistical Analyses

Exploratory factor analyses were performed on ADI-R items for both the ASD and ASD-negative groups. Specifically, the study used polychoric correlations between ADI-R items, the squared multiple correlation method for communality estimation, the principal factoring method for factor extraction, and the promax method of rotation. Retention of factors was guided by: (1) examination of the scree plot; (2) eigenvalues > 1.0 ; and (3) how easily factors could be interpreted based on item groupings. ADI-R items were determined to load onto a factor if the absolute value of the loading was ≥ 0.30 . Due to the limited number of RRB items rated as 2 or 3 (frequently or definitely observed) in the ASD-negative group,

it was necessary to collapse ratings of 1, 2, and 3 into one response category. Thus, for this group, each item was analyzed as a binary response, with 0 indicating behavior is never or rarely observed and 1 indicating behavior is occasionally or frequently observed. Furthermore, the ADI-R item of hand and finger mannerisms was excluded from the ASD-negative group exploratory factor analysis due to data sparsity and its collinearity with resistance to change in environment, although response frequencies for this item were still examined.

The relationship between participant characteristics and factor scores in the ASD group was examined using partial Pearson correlations, controlling for child age and sex. All analyses were conducted at the $\alpha = 0.05$ significance level using SAS version 9.3. For correlational analyses, RRB factor scores were examined in relation to individual characteristics identified from prior research, including communication and social features captured on the ADOS, behavioral symptoms from the CBCL, developmental levels related to receptive language, expressive language, and visual reception skills measured by the Mullen, and communication and social scores from the Vineland-II.

Results

ASD Group Exploratory Factor Analysis

Descriptive statistics for the 13 ADI-R items and response patterns found in the ASD group are displayed in Table 2. All children had at least one recorded value for one of the ADI-R items listed. Behaviors occurring at least occasionally for more than half of the participants with ASD included unusual sensory interests, repetitive use of objects or parts of objects, other complex mannerisms or stereotyped body movements, stereotyped utterances and delayed echolalia, hand and finger mannerisms, circumscribed interests, and difficulty with minor change in routines. Behaviors occurring at least occasionally for more than a quarter of participants included compulsions/rituals, abnormal response to sensory stimuli, unusual preoccupations, and verbal rituals. Behaviors with the fewest endorsements included resistance to change in environment and unusual attachment to objects.

The exploratory factor analysis for the ASD group retained three factors (Table 3). A repetitive sensorimotor behavior (RSMB) factor included unusual preoccupations (which includes repetitive or stereotyped interest in certain objects), repetitive use of objects or parts of objects, unusual sensory interests, hand and finger mannerisms, and other complex mannerisms or stereotyped body movements. The IS factor included compulsions/rituals, difficulty with minor change in routines, resistance to change in environment, and unusual attachment to objects (which may include an insistence on carrying around certain objects). Finally, a third repetitive stereotyped speech (SPEECH) factor included stereotyped utterances and delayed echolalia and verbal rituals. Circumscribed interests and abnormal response to specific sensory stimuli did not meet the 0.30 threshold for a high factor loading, but their highest loadings were both on SPEECH. The inter-factor correlation between RSMB and IS was 0.47, between RSMB and SPEECH was -0.06 , and between IS and SPEECH was 0.40.

A complete case analysis ($N = 637$) of partial correlations between participant characteristics and RRB factor scores, controlling for age and sex, is given in Table 4. RSMB was negatively correlated to a moderate degree with the Vineland-II communication and social composite scores (-0.45 and -0.46 , respectively, all $p < .01$) and modestly with Mullen visual reception, receptive language, and expressive language developmental age-equivalent scores (-0.39 , -0.40 , and -0.38 , respectively, all $p < .01$). RSMB also showed a modest positive correlation with the CBCL internalizing, ADHD, and externalizing scores (0.40 , 0.36 , and 0.36 , respectively, all $p < .01$) and the ADOS total social score (0.30 , $p < .01$). IS was positively correlated with the CBCL internalizing, externalizing, and anxiety T-scores to a modest degree (0.43 , 0.39 , and 0.33 , respectively, all $p < 0.01$).

SPEECH showed moderate positive correlations with the Mullen expressive language, receptive language, and visual reception age equivalent scores (0.47 , 0.43 , and 0.43 , respectively, $p < .01$) and the Vineland-II communication standard score (0.45 , $p < .01$). Interestingly, SPEECH did not correlate to the ADOS communication total score but did show a slight yet significant negative association with the ADOS social total score (-0.20 , $p < .01$). An item-specific analysis of partial Pearson correlations between each individual subject characteristic and RRB factor scores did not substantively alter the direction or significance level of the results compared to the complete case analysis.

ASD-negative Group Exploratory Factor Analysis

Descriptive statistics for the 13 ADI-R items considered for the ASD-negative group analysis are displayed in Table 5. Compared to the ASD group, fewer ADI-R RRB items were endorsed across items. Items most frequently endorsed (occurring at least occasionally in about 27 to 40 percent of this group) included unusual sensory interests, repetitive use of objects or parts of objects, stereotyped utterances and delayed echolalia, difficulty with minor changes in routine, and circumscribed interests, although these were all reported to a lesser degree compared to the ASD group.

The ASD-negative group exploratory factor analysis also retained three factors (Table 6), though factor loadings were inconsistent and difficult to interpret. Factor 1 included unusual preoccupations, compulsions/rituals, difficulty with minor change in routines, and resistance to change in environment, perhaps resembling an insistence on sameness factor. Factor 2 included stereotyped utterances and delayed echolalia, repetitive use of objects or parts of objects, unusual sensory interests, abnormal response to specific sensory stimuli, and unusual attachment to objects, along with verbal rituals which had a higher loading on factor 3. Factor 3 included verbal rituals, unusual attachment to objects, and other complex mannerisms or stereotyped body movements. Circumscribed interests did not meet the 0.30 threshold for a high factor loading, but its highest loading was on Factor 2. Inter-factor correlations between Factor 1 and Factor 2 was 0.41, between Factor 1 and Factor 3 was 0.04, and between Factor 2 and Factor 3 was 0.11.

Discussion

This analysis is among the first to examine RRBs and their factor structure among a large community-based sample of preschool-aged children with confirmed autism (ASD

group) alongside a group of children with non-ASD developmental concerns (ASD-negative group). For this analysis, a wide range of ADI-R RRB items were included to reflect ASD diagnostic conceptualizations described in DSM-5. Given that developmental changes occur quite rapidly for young children, it is important to examine symptom presentation at specific age ranges within the early developmental period. In previous factor analysis studies focusing on young children including those under 3 years of age, certain RRB items related to circumscribed interests and repetitive language features were omitted due to limited language among participants and age restrictions for item administration on the ADI-R. The current analysis focused exclusively on preschool-age children, primarily between ages 3 to 5 years; thus, for this sample, circumscribed interest and repetitive speech items were retained and yielded different factor structure findings. Similar to other studies examining children with ASD diagnoses at younger ages, this ASD sample contained a large majority of males, with individuals in the ASD group showing greater developmental delays in terms of visual reception/nonverbal problem-solving, receptive language, expressive language, and adaptive skills compared to the ASD-negative group.

As expected, individuals in the ASD group exhibited high rates of multiple RRB symptoms. Descriptively, the pattern of RRBs between the ASD group and ASD-negative group were similar in that the most widely endorsed RRBs included unusual sensory interests, repetitive use of objects or parts of objects, and stereotyped utterances and delayed echolalia suggesting that these may be the most common RRBs among this age group, though these were more frequent among the ASD group. Behaviors that were less commonly reported across both groups in this age range included resistance to change in environment and unusual attachment to objects. Findings related to the most common endorsements within the ASD group are consistent with prior literature examining RRBs endorsed among young children with autism-related diagnoses. For example, Richler et al. (2007) found that unusual sensory interests, repetitive object use, complex mannerisms, and hand/finger mannerisms were common and endorsed in over half of their sample of children with ASD at 2 years of age. Such findings for these specific behaviors held true for the preschool-aged children in the present analysis.

Notably, complex mannerisms and hand and finger mannerisms were widely endorsed in the ASD group (present for 54.5% and 53.5% of participants, respectively), and much less so in the ASD-negative group (present for 16.7% and 8.3%, respectively) indicating these behaviors are not as prevalent for children with non-autism developmental delays in this age range. In addition, circumscribed interest and stereotyped utterance items included in this analysis were also fairly common and reported to some degree in more than half of individuals in the ASD group, with nearly a third or more children showing more definite/interfering symptoms within these areas (30.4% for circumscribed interests and 35.2% for stereotyped utterances). In contrast, the majority of children in the ASD-negative group did not have any behaviors endorsed within these areas, and very few showed symptoms that were definite or interfering (6.6% and 4.2%, respectively). Such items could prove to be important in differentiating preschool-age children with ASD from those with other developmental concerns.

Given the study design and sample selection methods, it was expected that RRB factor structures would differ between the ASD and ASD-negative groups; however, it is interesting to qualitatively consider the differences in how RRB items grouped. Results of the exploratory factor analysis for the ASD group yielded a well-defined three-factor structure that provided a good fit to the data and resulted in ADI-R item groupings that were easily interpretable. However, the three-factor structure that emerged for the ASD-negative group was difficult to conceptualize, in part due to the limited item endorsements and the need to collapse response options for this group. One possible reason for findings in the ASD-negative group is the potential for measurement invariance in RRBs across clinical populations. Evaluation of the psychometric properties of RRB measures is needed to ensure that interpretation of findings is similar in children with and without ASD.

For the ASD group, items relating to stereotyped utterances and delayed echolalia as well as verbal rituals comprised a Stereotyped Speech (SPEECH) factor. Behaviors captured within this factor include repetitive and nonsocial speech such as self-commentary, scripting of prior inter-changes, and routinized phrases, as well as compulsive repetitive speech or insistence on others saying something repeatedly. An Insistence on Sameness (IS) factor also emerged related to difficulties with minor changes in routines, resistance to change in the environment, unusual attachment to items, and compulsions/rituals, in addition to a Repetitive Sensorimotor Behaviors (RSMB) factor characterized by repetitive use of actions, unusual sensory interests, hand/finger mannerisms, complex/stereotyped body mannerisms, and unusual preoccupations. The latter two factors have been repeatedly reported in the literature (Berry et al. 2018).

The emergence of a distinct three-factor structure of RRBs for the ASD group including a novel SPEECH factor, which has not yet been described in the current literature, highlights the importance of including RRB items that capture the full range of behaviors described in current ASD diagnostic criteria and may add evidence that behavioral symptom presentation may be unique to the stage of development during which participants are assessed. A SPEECH factor may represent a more clinically relevant symptom domain among preschool-aged children since increased expressive language skills developed during preschool years may be accompanied by emergence of unusual speech patterns. In contrast to previous three-factor models found among older age ranges and in line with findings for younger children, a distinct circumscribed interest (CI) factor did not emerge for the preschool-age ASD group, as circumscribed interests have been associated with higher developmental level and may increase in severity with age (South et al. 2005). Further, the circumscribed interest item has typically been excluded for children under 3 years of age when using the ADI-R. Unusual preoccupations, which has loaded on a CI factor in some previous studies of older children, instead loaded on the RSMB factor in our study of preschool children. This finding could indicate that unusual interests serve different developmental functions at different ages. While the factor structure that emerged in this study does not necessarily mirror the conceptually-driven RRB categories currently described in DSM-5, which aim to describe autism-related behaviors that may be apparent at any point in development, current findings highlight features that might be particularly relevant to monitor during the preschool years.

Although RRB factors found among the ASD group were slightly correlated with each other, they generally were associated with separate clinical correlates, adding support to these factors as distinct behavioral features. Differing associations between RRBs and clinical characteristics have been found for distinct RRB subtypes across prior studies (Bruckner and Yoder 2007; Harrop et al. 2014; Honey et al. 2007; Joseph et al. 2013; Mahone et al. 2004). For example, some studies have reported that “lower-order” RRBs, including repetitive use of objects, hand and finger mannerisms, and some sensory behaviors, are associated with younger age, lower nonverbal cognitive functioning, decreased adaptive skills, and more severe social communication deficits, while “higher-order” RRBs, such as insistence on sameness, ritualistic behaviors, and circumscribed interests are more prevalent in older children with ASD and are associated with higher nonverbal cognitive levels (Baranek et al. 2007; Bishop et al. 2006; Lam et al. 2008; Richler et al. 2010; South et al. 2005).

Similar to prior studies, RSMB for the ASD group in this analysis was associated with lower developmental and adaptive functioning. In addition, RSMB was linked to increased social challenges as measured by the ADOS, and other emotional and behavioral concerns; specifically, internalizing, externalizing, ADHD-related, and emotion regulation difficulties measured on the CBCL, suggesting that higher levels of RSMB symptoms may predict greater levels of broader behavioral concerns. IS showed the strongest positive relationship with internalizing scores on the CBCL, possibly due to an association with internalizing symptoms such as anxiety (also associated with IS to a moderate degree), in line with prior findings linking insistence on sameness to increased anxiety (Gotham et al. 2013; Lidstone et al. 2014). There was also a modest positive association with externalizing symptoms, possibly related to behavioral challenges linked to insistence on sameness difficulties. Similar to findings from Bishop et al. (2006), IS was not correlated with child developmental level in this sample.

While others have found that behavioral features such as stereotyped utterances/delayed echolalia and verbal rituals are linked to lower developmental functioning among school-age children (Bishop et al. 2006), the SPEECH factor in this sample showed the strongest positive associations with expressive and receptive language levels and visual reception scores on the Mullen as well as adaptive communication scores on the Vineland-II. For preschool-age children, this association may be in line with the assumption that atypical speech symptoms emerge with the development of basic language skills that provide a basis for echoed, scripted, or repetitive speech in young children, while the persistence of such speech patterns in samples of older children might suggest more limitations in flexible language use and general developmental functioning. SPEECH was not correlated to ADOS communication total score, likely due to this score capturing functional communication used for social purposes, which would not necessarily be linked to presence of stereotyped or repetitive speech patterns lacking social intent.

Caregiver report on the ADI-R was used to examine RRBs given that this tool is considered one of the gold-standard measures of autism symptoms that captures a broad range of relevant behavioral features. Although reliance on caregiver report to assess behaviors has been noted to be a limitation within research, ADI-R ratings provide an advantage for capturing behaviors witnessed by parents that may not have been directly observed in

time-limited assessments administered directly to children by clinicians. It is also important to note that separate ADI-R algorithms have been proposed for toddlers and young children 12 to 47 months of age (Kim and Lord 2012). These algorithms include items other than those used in this study given differential manifestations of ASD at different ages. The mean age of children with ASD in our sample was 59 months and only 6% were younger than 47 months when the ADI-R was administered. Nonetheless, future analyses should consider differences between ADI-R toddler and school-aged algorithms when examining the factor structure of ASD symptoms.

In terms of study limitations, the use of a single measure such as the ADI-R may still underrepresent the full range of RRBs an individual may exhibit (Lecavalier et al. 2006); thus, additional measures to assess RRB symptoms should continue to be used to explore latent RRB constructs that are not specific to the ADI-R. Additionally, this analysis included a large number of items relative to sample size, particularly among the ASD-negative comparison group. Collapsing ratings for this group also weakened the comparison of factor structures between groups. Future analyses should examine RRBs among larger samples of individuals with confirmed ASD, non-ASD developmental concerns, and children who are typically developing to provide better cross-comparisons.

While this analysis focused on an exploratory factor analysis, a logical next step would be confirmatory factor analysis to determine if this novel three-factor structure would be found in another large sample of preschool-age children with ASD. Further, it would be interesting to further examine whether such a factor structure would remain for children within narrower age and developmental/IQ ranges within the preschool period, as some longitudinal studies have shown that RRB patterns may shift for individuals over relatively short periods (Honey et al., 2008). Finally, although prior analyses have excluded atypical speech items from RRB analyses using the ADI-R given that these are not administered to children under age 3 years, more research is needed to fully pinpoint the emergence and trajectory of atypical speech patterns at earlier age ranges such as atypical babbling, use of jargon-like sounds, and repetitive word use that children may exhibit at younger ages before fully functional phrase speech has been established.

Ultimately, findings from this analysis refine our understanding of RRB subtypes among preschool-age children with ASD. Due to the complexity of ASD diagnostics, it is important to differentiate RRB patterns that may be specific to ASD versus behaviors that are observed in other populations of young children, including those with other neurodevelopmental concerns. Furthermore, because RRBs present from a very young age, better understanding these symptoms could aid earlier ASD diagnosis and lead to earlier delivery of targeted intervention services. The possibility that RRB factor structures differ depending on very specific age ranges highlights the importance of further research to examine possible underlying neurobiological and developmental drivers affecting RRB symptom presentation over the course of development and the need for more longitudinal research examining symptoms trajectory at specific periods of development.

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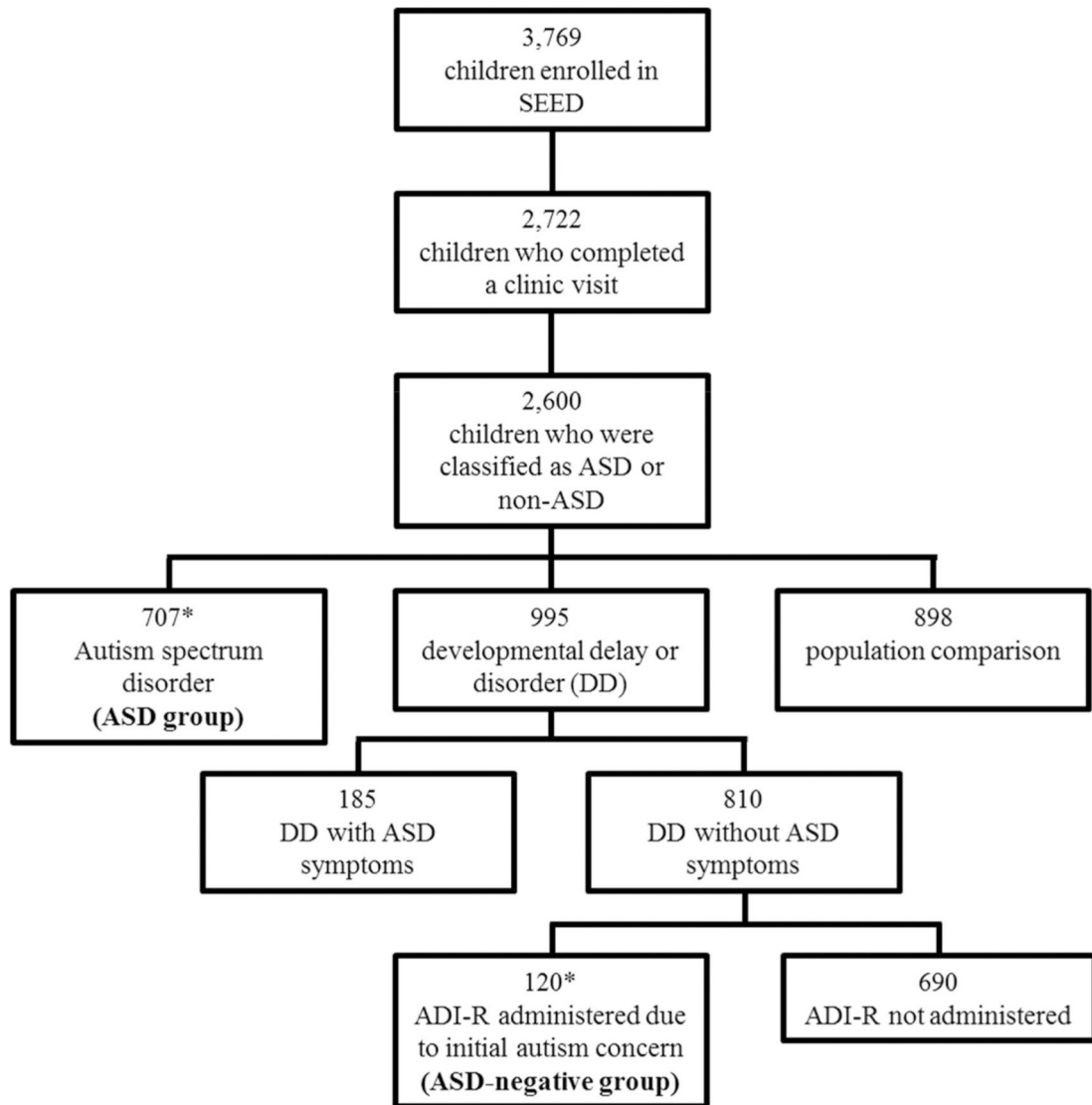
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* Included in analysis

Fig. 1. Flow chart for SEED-enrolled children selected for this study’s ASD and ASD-negative groups

Table 1

Descriptive characteristics of children in ASD and ASD-negative groups

| | Case status * | | p-value ** | N missing |
|---|----------------|-------------------------|------------|-----------|
| | ASD N = 707 | ASD-negative N = 120 | | |
| Demographics | | | | |
| Males | 580 (82%) | 81 (68%) | < .001 | |
| Maternal race | | | < .001 | |
| White | 481 (68%) | 66 (55%) | | |
| Black | 152 (21%) | 46 (38%) | | |
| Other, multiracial | 71 (10%) | 5 (4%) | | |
| Unknown | 3 (0%) | 3 (3%) | | |
| Maternal ethnicity | | | 0.008 | |
| Non-Hispanic | 533 (75%) | 80 (67%) | | |
| Hispanic | 76 (11%) | 23 (19%) | | |
| Unknown | 98 (14%) | 17 (14%) | | |
| Maternal education | | | < .001 | |
| 8th grade or less | 11 (2%) | 9 (8%) | | |
| 9th–12th grade, no diploma | 27 (4%) | 17 (14%) | | |
| High school graduate or general education development certificate | 121 (17%) | 30 (25%) | | |
| Some college, no degree | 164 (23%) | 28 (23%) | | |
| Bachelor's degree | 177 (25%) | 17 (14%) | | |
| Advanced degree | 194 (27%) | 17 (14%) | | |
| Unknown | 13 (2%) | 2 (2%) | | |
| Child age (months) | 59.3 ± 6.7 | 60.2 ± 7.6 | 0.22 | 4 |
| Clinical characteristics | | | | |
| SCQ total score | 17.4 ± 6.1 | 13.8 ± 3.1 | < .001 | 3 |
| ADI-R | | | | |
| Reciprocal social interaction total | 18.3 ± 5.9 | 5.7 ± 4.3 | < .001 | 0 |
| Communication total (verbal) | 14.7 ± 4.2 | 6.0 ± 3.9 | < .001 | 211 |
| Communication total (nonverbal) | 11.4 ± 2.4 | 2.8 ± 2.0 | < .001 | 616 |
| Restricted/repetitive behaviors total | 6.0 ± 2.6 | 2.3 ± 1.9 | < .001 | 0 |

| | Case status* | | p-value** | N missing |
|--|----------------|-------------------------|-----------|-----------|
| | ASD N = 707 | ASD-negative N = 120 | | |
| ADOS | | | | |
| Communication total | 5.2 ± 1.9 | 1.4 ± 1.2 | < .001 | 0 |
| Reciprocal social interaction total | 9.0 ± 2.8 | 1.9 ± 1.5 | < .001 | 0 |
| Stereotyped behaviors/restricted interests total | 3.3 ± 1.7 | 1.0 ± 1.1 | < .001 | 0 |
| CBCL | | | | |
| Attention deficit and hyperactivity T-score | 60.4 ± 8.2 | 57.6 ± 8.0 | < .001 | 58 |
| Affective T-score | 61.9 ± 10.0 | 59.4 ± 9.1 | 0.013 | 58 |
| Anxiety T-score | 57.5 ± 9.3 | 59.6 ± 10.6 | 0.028 | 58 |
| Externalizing T-score | 60.3 ± 11.4 | 57.1 ± 12.1 | 0.006 | 58 |
| Internalizing T-score | 62.5 ± 9.6 | 58.2 ± 11.3 | < .001 | 58 |
| Mullen | | | | |
| Expressive language age equivalent | 35.0 ± 16.3 | 47.7 ± 12.7 | < .001 | 10 |
| Receptive language age equivalent | 37.3 ± 17.9 | 49.2 ± 12.1 | < .001 | 11 |
| Visual reception age equivalent | 43.3 ± 16.9 | 53.1 ± 10.4 | < .001 | 5 |
| Early learning composite standard score | 66.9 ± 20.0 | 79.6 ± 17.5 | < .001 | 12 |
| Vineland-II | | | | |
| Communication standard score | 77.0 ± 18.2 | 87.5 ± 12.6 | < .001 | 4 |
| Socialization standard score | 72.6 ± 13.4 | 89.3 ± 11.7 | < .001 | 6 |
| Adaptive behavior composite standard score | 75.7 ± 28.2 | 96.4 ± 51.9 | < .001 | 7 |

SCQ Social Communication Questionnaire, ADI-R Autism Diagnostic Interview-Revised, ADOS Autism Diagnostic Observation Schedule, CBCL Child Behavior Checklist 1½–5, Mullen Mullen Scales of Early Learning, Vineland-II Vineland Adaptive Behavior Scales-Second Edition

* Values expressed as n (%) or mean ± standard deviation

** P value comparisons between case status are based on chi-square test of homogeneity for categorical variables (unknown categories are excluded from the test); P values for continuous variables are based on ANOVAV

Table 2

ADI-R response frequencies in ASD group

| Item no | Item descriptor | N | Frequency of 0, 1, 2, or 3 Responses in %* | | | | Mean (SD) |
|---------|--|-----|--|------|------|------|-------------|
| | | | 0 | 1 | 2 | 3 | |
| 33 | Stereotyped utterances delayed echolalia | 707 | 42.0 | 22.8 | 34.1 | 1.1 | 0.94 (0.90) |
| 39 | Verbal rituals | 706 | 71.2 | 13.5 | 12.5 | 2.8 | 0.47 (0.82) |
| 67 | Unusual preoccupations | 705 | 67.7 | 13.8 | 11.6 | 7.0 | 0.58 (0.95) |
| 68 | Circumscribed interests | 703 | 46.7 | 22.9 | 20.3 | 10.1 | 0.94 (1.04) |
| 69 | Repetitive use of objects or parts of objects | 706 | 28.5 | 31.3 | 24.2 | 16.0 | 1.28 (1.04) |
| 70 | Compulsions/rituals | 705 | 56.6 | 15.7 | 17.4 | 10.2 | 0.81 (1.06) |
| 71 | Unusual sensory interests | 705 | 26.2 | 44.5 | 29.2 | N/A | 1.03 (0.74) |
| 73 | Abnormal response to sensory stimuli | 704 | 63.4 | 19.9 | 12.6 | 4.1 | 0.58 (0.86) |
| 74 | Difficulty with minor change in routines | 705 | 48.1 | 24.0 | 21.1 | 6.8 | 0.87 (0.97) |
| 75 | Resistance to change in environment | 704 | 86.9 | 7.2 | 4.3 | 1.6 | 0.20 (0.58) |
| 76 | Unusual attachment to objects | 702 | 76.4 | 12.0 | 9.7 | 2.0 | 0.37 (0.74) |
| 77 | Hand and finger mannerisms | 703 | 46.5 | 18.9 | 27.6 | 7.0 | 0.95 (1.01) |
| 78 | Other complex mannerisms or stereotyped body movements | 704 | 45.5 | 19.6 | 26.4 | 8.5 | 0.98 (1.03) |

* Note: SD = standard deviation

0 responses generally indicate the behavior is never or rarely observed

1 responses generally indicate the behavior is occasionally/mildly observed

2 responses generally indicate the behavior is frequently observed/definite and may cause some interference

3 responses generally indicate the behavior is frequently observed/definite and interfering NA indicates this rating does not apply for this item

Table 3

Promax rotated factor pattern of ASD group exploratory analysis with 13 ADI-R items (N = 702)

| Item no | Item descriptor | R SMB | IS | SPEECH |
|---------|--|--------------|--------------|--------------|
| 33 | Stereotyped utterances and delayed echolalia | -0.021 | -0.089 | 0.716 |
| 39 | Verbal rituals | -0.019 | 0.188 | 0.615 |
| 67 | Unusual preoccupations | 0.358 | 0.042 | 0.054 |
| 68 | Circumscribed interests | 0.083 | 0.204 | <u>0.260</u> |
| 69 | Repetitive use of objects or parts of objects | 0.601 | 0.172 | -0.053 |
| 70 | Compulsions/rituals | -0.025 | 0.641 | 0.169 |
| 71 | Unusual sensory interests | 0.539 | 0.084 | -0.035 |
| 73 | Abnormal response to specific sensory stimuli | 0.214 | 0.089 | <u>0.270</u> |
| 74 | Difficulty with minor change in routines | -0.007 | 0.629 | 0.118 |
| 75 | Resistance to change in environment | 0.011 | 0.660 | 0.042 |
| 76 | Unusual attachment to objects | 0.192 | 0.411 | -0.168 |
| 77 | Hand and finger mannerisms | 0.637 | -0.124 | 0.027 |
| 78 | Other complex mannerisms or stereotyped body movements | 0.599 | -0.024 | 0.108 |

R SMB repetitive sensorimotor behaviors, *IS* insistence on sameness, *SPEECH* stereotyped speech Bold numbers indicate factor loadings > 0.30, underlined numbers represent the highest factor loading (< 0.30) for the item

Table 4

Partial Pearson correlations between ASD group subject characteristics and RRB factor scores for 13 ADI-R items (N = 637)

| | Pearson correlations | | |
|---|----------------------|---------|---------|
| | RSMB | IS | SPEECH |
| ADOS communication total | 0.20** | 0.06 | 0.01 |
| ADOS social total | 0.30** | 0.01 | -0.20** |
| CBCL ADHD T-score | 0.36** | 0.29** | 0.05 |
| CBCL affective T-score | 0.33** | 0.29** | 0.02 |
| CBCL anxiety T-score | 0.21** | 0.33** | 0.18** |
| CBCL externalizing T-score | 0.36** | 0.39** | 0.12** |
| CBCL internalizing T-score | 0.40** | 0.43** | 0.15** |
| Mullen expressive language age equivalent | -0.38** | 0.05 | 0.47** |
| Mullen receptive language age equivalent | -0.40** | 0.01 | 0.43** |
| Mullen visual reception age equivalent | -0.39** | 0.01 | 0.43** |
| Vineland-II communication standard score | -0.45** | -0.03 | 0.45** |
| Vineland-II social standard score | -0.46** | -0.15** | 0.28** |

ADOS Autism Diagnostic Observation Schedule, *CBCL* Child Behavior Checklist 1½–5, *Mullen* Mullen Scales of Early Learning, *Vineland-II* Vineland Adaptive Behavior Scales-Second Edition, *AE* age equivalent, *IS* insistence on sameness, *RSMB* repetitive sensorimotor behavior, *SPEECH* stereotyped speech

**
p < 0.01

Table 5

ADI-R response frequencies in ASD-negative group

| Item no | Item descriptor | N | Frequency of 0, 1, 2, or 3 responses in %* | | | | Mean (SD) |
|---------|--|-----|--|------|-----|-----|-------------|
| | | | 0 | 1 | 2 | 3 | |
| 33 | Stereotyped utterances and delayed echolalia | 120 | 65.0 | 30.8 | 4.2 | 0.0 | 0.39 (0.57) |
| 39 | Verbal rituals | 120 | 87.5 | 10.0 | 2.5 | 0.0 | 0.15 (0.42) |
| 67 | Unusual preoccupations | 120 | 91.7 | 5.8 | 2.5 | 0.0 | 0.11 (0.38) |
| 68 | Circumscribed interests | 120 | 72.5 | 20.8 | 5.8 | 0.8 | 0.35 (0.63) |
| 69 | Repetitive use of objects or parts of objects | 120 | 64.2 | 30.0 | 5.8 | 0.0 | 0.42 (0.60) |
| 70 | Compulsions/rituals | 120 | 84.2 | 9.2 | 6.7 | 0.0 | 0.23 (0.56) |
| 71 | Unusual sensory interests | 120 | 60.0 | 35.0 | 5.0 | N/A | 0.45 (0.59) |
| 73 | Abnormal response to specific sensory stimuli | 120 | 84.2 | 11.7 | 4.2 | 0.0 | 0.20 (0.50) |
| 74 | Difficulty with minor change in routines | 120 | 70.8 | 20.8 | 7.5 | 0.8 | 0.38 (0.66) |
| 75 | Resistance to change in environment | 120 | 91.7 | 5.8 | 2.5 | 0.0 | 0.11 (0.38) |
| 76 | Unusual attachment to objects | 120 | 86.7 | 6.7 | 6.7 | 0.0 | 0.20 (0.54) |
| 77 | Hand and finger mannerisms | 120 | 91.7 | 5.8 | 2.5 | 0.0 | 0.11 (0.38) |
| 78 | Other complex mannerisms or stereotyped body movements | 120 | 83.3 | 12.5 | 3.3 | 0.8 | 0.22 (0.54) |

* Note SD = standard deviation

0 responses generally indicate the behavior is never or rarely observed

1 responses generally indicate the behavior is occasionally/mildly observed

2 responses generally indicate the behavior is frequently observed/definite and may cause some interference

3 responses generally indicate the behavior is frequently observed/definite and interfering NA indicates this rating does not apply for this item

Table 6
Promax rotated factor pattern of ASD-negative group exploratory analysis with 12 ADI-R items (N = 120)

| Item No | Item Descriptor | Factor 1 | Factor 2 | Factor 3 |
|---------|--|--------------|--------------|---------------|
| 33 | Stereotyped utterances and delayed echolalia | 0.067 | 0.424 | 0.135 |
| 39 | Verbal rituals | - 0.057 | 0.411 | 0.485 |
| 67 | Unusual preoccupations | 0.606 | - 0.078 | - 0.096 |
| 68 | Circumscribed interests | 0.085 | <u>0.252</u> | - 0.023 |
| 69 | Repetitive use of objects or parts of objects | 0.045 | 0.644 | 0.16 |
| 70 | Compulsions/rituals | 0.769 | - 0.083 | 0.231 |
| 71 | Unusual sensory interests | 0.089 | 0.768 | 0.072 |
| 73 | Abnormal response to specific sensory stimuli | - 0.191 | 0.654 | - 0.182 |
| 74 | Difficulty with minor change in routines | 0.691 | 0.186 | 0.059 |
| 75 | Resistance to change in environment | 0.89 | 0.042 | - 0.207 |
| 76 | Unusual attachment to objects | 0.085 | 0.417 | - 0.64 |
| 78 | Other complex mannerisms or stereotyped body movements | 0.028 | 0.154 | 0.734 |

Note: bold numbers indicate factor loadings ≥ 0.30 or ≤ -0.30 , underlined numbers represent the highest factor loading (< 0.30) for the item