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Patterns of Restricted and Repetitive Behaviors in Toddlers and Young Children with Autism Spectrum Disorder

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Objectives: Restricted and repetitive behaviors (RRBs) are a core symptom in the diagnosis of autism spectrum disorder (ASD). The complexity of behavioral patterns has called for the creation of phenotypically homogeneous subgroups among individuals with ASD. The purpose of this study was 1) to investigate the different types of RRBs and 2) to explore whether subgroups created by RRBs would show unique levels of functioning in toddlers and young children with ASD.
Methods: A total of 313 children with ASD, aged 12–42 months were included in the analysis. The Autism Diagnostic Interview-Revised was used to obtain information on the different types of RRBs by grouping 15 items into six categories. The Vineland Adaptive Behaviors Scale, a parent-reported questionnaire, was used to measure adaptive functioning. A portion of the children were analyzed separately for verbal-related RRBs based on their expressive language level. Two-step cluster analysis using RRB groups as features was used to create subgroups. Analysis of covariance while covarying for age and language was performed to explore the clinical characteristics of each cluster group.
Results: Sensory-related RRBs were the most prevalent, followed by circumscribed interests, interest in objects, resistance to change, and repetitive body movements. A subset of the children was analyzed separately to explore verbal-related RRBs. Four cluster groups were created based on reported RRBs, with multiple RRBs demonstrating significant delays in adaptive functioning.
Conclusion: Heterogeneity of RRBs emerges at a young age. The different patterns of RRBs can be used as valuable information to determine developmental trajectories with better implications for treatment approaches.

Keywords: Autism spectrum disorder; Restricted and repetitive behaviors; Cluster analysis; Adaptive functioning.

INTRODUCTION

Restricted and repetitive behaviors (RRBs) constitute a core symptom domain of autism spectrum disorder (ASD), a behaviorally defined neurodevelopmental condition. With an estimated prevalence of 1 in 44 children [1], ASD diagnoses have been made based on the presentation of impaired social communication and RRBs [2]. RRBs represent a broad spectrum of responses that encompass stereotyped motor mannerisms, non-typical sensory interests, repetitive use of objects, preoccupation with unusual objects, adherence to non-functional routines, or interest in objects with abnormal intensity and circumscribed nature [3]. These topographies manifest differently in individuals with ASD, and can change in quantity, quality, and type over time. More importantly,

engagement in RRBs can interfere with opportunities for children to develop adaptive skills [4], interact socially with others [5], and engage in daily activities.

Hence, to better capture the heterogeneous nature of RRBs, a two-factor model that clusters behaviors as either lower or higher order has been widely applied [6]. Lower-order RRBs focus on stereotypes and repetitive sensorimotor behaviors, whereas higher-order RRBs refer to adherence to rituals or routines, resistance to change, and insistence on sameness. Other studies have further suggested additional subcategories, such as self-injurious behaviors (e.g., head banging, skin picking, face slapping, excessive self-rubbing, self-biting) [7], and circumscribed interests (e.g., perseveration on a particular topic, object, or theme) [8]. However, depending on the assessment instrument used to measure behaviors and the age of individuals with ASD, previous findings have reported between two and five RRB subtypes [6,9,10].

The categorization of RRBs has helped unravel meaning-

ful behavioral patterns by underscoring the criticality of the early diagnosis of RRBs and their strong associations with cognitive functioning [10,11], co-occurring psychiatric symptoms [12], and adaptive behaviors [13]. For example, findings report that the prevalence of RRBs is negatively associated with the intelligence quotient (IQ) among individuals with ASD. More specifically, these associations can vary depending on the RRB type. Individuals with higher cognitive functioning exhibit a greater frequency of higher-order RRBs [11], and those with lower cognitive ability have more severe lower-order RRBs [10]. Stratis and Lecavlier [12] found that different types of RRBs are predictive of anxiety, depression, and oppositional defiant disorder-related symptoms. Others emphasized the need for interventions targeting RRBs to enhance overall functioning of individuals with ASD and found a negative association between the two [13,14]. Therefore, the degree to which RRBs can affect various functioning skills and general well-being in children with ASD underlines the importance of understanding this wide-ranging class of behaviors.

Although RRBs possess strong diagnostic significance within ASD, little is known about the types of RRBs and their association with other developmental domains in toddlers and young children. RRBs can typically be identified as early as 12 months [15]. While previous studies have proposed that RRBs are likely constructive factors of early child development [16], discerning RRBs unique to ASD from normative RRBs has been challenging for researchers. Though some RRBs may be specific to ASD, most topographies of repetitive behavior are observed in children without ASD at some stage during their developmental course [17]. Ultimately, it is imperative to resolve the ambiguity behind distinguishing RRBs that denote risk of ASD. This study aimed to investigate RRB subtypes and explore whether subgroups created by theses behaviors show unique levels of functioning in toddlers and young children with ASD. While we considered the analysis of the prevalence of RRB subtypes to be exploratory, we hypothesized that children with lower-order RRBs would have more difficulties in adaptive skills.

METHODS

Participants

This was a retrospective study with data acquired from two larger projects (i.e., validating a novel ASD screening instrument and detecting early risk signs of ASD using artificial intelligence). Participants were recruited through posters at Seoul National University Bundang Hospital outpatient clinics, local healthcare centers, and online communities. The Institutional Review Board of Seoul National University Bun-

dang Hospital approved the use of fully anonymized data (IRB no: B-2112-729-103).

Questionnaires regarding developmental history, language ability, autistic traits, and adaptive behavior were mailed and collected on-site during the participants' visit. Gold-standard diagnostic evaluations were administered by trained researchers, and each assessment was video-recorded and reviewed to ensure quality and rater consensus. The child's final diagnosis was based on the best-estimate diagnosis by licensed child psychiatrists, clinical psychologists, and special education professionals. Only those who met the Diagnostic and Statistical Manual of Mental Disorders (5th edition) [2] ASD criteria were included in this study.

Measurements

Autism Diagnostic Interview-Revised (ADI-R)

The ADI-R [18] is a structured caregiver interview used to diagnose ASD. It comprises 93 question items rated on the child's current or past behavior. Scoring is based on an ordinal scale with higher numbers indicating greater symptom severity. Specific items are grouped into the following four domains, each with a cut-off score: 1) reciprocal social interaction, 2) communication, 3) RRBs, and 4) presence of abnormal behavior at or before 36 months. The Korean translated version approved by the Western Psychological Services (WPS) was used [19].

Autism Diagnostic Observation Schedule (ADOS-2)

The ADOS-2 [20] is a structured interactive assessment used to diagnose ASD based on current behavior and skillsets. There are five modules contingent on the participants' chronological age, developmental level, and expressive verbal abilities. The toddler module (n=93), module 1 (n=190), and module 2 (n=30) were administered. The diagnostic algorithm consists of domains (i.e., social affect, RRB), which can be computed to produce a total score. Each module provides a different range for the total score to be classified as autism, autism spectrum, or non-autism. Calibrated severity scores were used to compare different modules. The WPS-approved Korean version of the ADOS-2 was used [21].

Vineland Adaptive Behavior Scale, Second Edition (VABS-II)

The VABS-II, a caregiver-report questionnaire, was used to measure adaptive functioning [22]. Caregivers were asked to score how frequently their child could execute the given items, with higher scores indicating consistent and generalized use. The subdomain scores were then compiled into four domains: 1) communication, 2) daily life, 3) socialization, and

4) motor skills. The Adaptive Behavior Composite, based on the four domains, calculates a standard total score provided on an age-matched norm sample. Standardized scores can be divided into five qualitative descriptors (20–70 low, 71–85 moderately low, 86–114 adequate, 115–129 moderately high, 130–140 high).

Other questionnaires

Other questionnaires, such as the Korean translated Social Responsiveness Scale [23] and the Social Communication Questionnaire [24], were completed by the caregivers and used to measure autistic traits. Higher scores indicated greater symptom severity for both instruments. Additionally, caregivers completed the Sequenced Language Scale for Infants (SELSI) [25] regarding their children's receptive and expressive language development. Age-matched developmental quotients and scores were used to determine potential language delays. Data collected from these instruments were used to provide additional information about each child and was used to obtain the best-estimate diagnosis.

Data analyses

The prevalence of RRBs was explored using 15 ADI-R items grouped into six categories adapted from Huerta et al. [26] (Table 1). Based on the suggestions of Leekam et al. [3], repetitive motor behaviors were split between repetitive body movements and repetitive behaviors with objects. Due to the imbalance of items per category, each group was binarily coded as present if caregivers reported at least one current behavior among the grouped ADI-R items, regardless of severity. A subset of children was then analyzed separately for verbal-related RRBs based on their expressive language ability. Lastly, a two-step cluster analysis, which uses a log-likelihood distance measure based on categorical or continuous variables to find the optimal subgroup model [27], created clusters using binary coding of the six RRB categories. Univariate analysis of covariance, while covarying for age and expressive language usage, was performed to explore the specific clinical characteristics of each group. Usage of expressive language was coded as a binary variable using the ADI-R item 30. All pairwise comparisons were Bonferroni-adjusted to account for multiple comparisons. Statistical analyses were conducted using SPSS version 26.0 (IBM Corp., Armonk, NY, USA).

RESULTS

In total, 313 children, predominantly male (79.6%) and aged 12–42 months (M=33.39, SD=6.71), were included in the analysis. Based on the ADI-R RRB item grouping, sensory-relat-

Table 1. Groupings of the ADI-R RRB items

RRB category	ADI-R item
Object interests	69. Repetitive use of objects or interest in parts of objects
Resistance to change	70. Compulsions/rituals
	74. Difficulties with minor changes in own routines or personal environment
	75. Resistance to trivial changes in environment
Restricted interest	67. Unusual preoccupations
	68. Circumscribed interests
	76. Unusual attachment
Sensory interests	71. Unusual sensory interests
	72. Sensitivity to noise
	73. Sensory aversions
Body movements	77. Hand or finger mannerisms
	78. Other complex mannerisms or stereotyped body movements
	79. Complex mannerisms
Verbal	33. Stereotyped utterances and delayed echolalia
	39. Verbal rituals

Only a subset of the children was analyzed for the verbal RRB category. RRB, restricted and repetitive behavior; ADI-R, Autism Diagnostic Interview-Revised

ed RRBs (71.9%) were the most prevalent, followed by circumscribed interest (61%), interest in objects (58.1%), resistance to change (47%), and repetitive body movements (42.8%). Of the 69 children who met the minimum language requirement, 52 (75.3%) reported verbal-related RRBs.

The two-step cluster analysis resulted in four subgroups (Table 2). Cluster 1 (n=87) included all non-verbal children with sensory and object-related RRBs, thus labeled 'multiple RRBs'. Cluster 2 (n=102) and cluster 3 (n=73) were respectively considered 'resistance to change RRBs' and 'sensory RRBs,' given that all the children in each group reported the presence of the outlined behavior. Finally, cluster 4 (n=51) was the smallest group but was classified as the 'least RRBs' because no children in this group had either resistance to change or sensory interest RRBs.

Clinical assessment score comparisons between the cluster groups showed that symptom severity was greatest in cluster 1, relatively mild in cluster 3, and least measured in clusters 2 and 4 (Table 3). Bonferroni-adjusted pairwise comparisons indicated that the most prominent difference was observed between clusters 1 and 2.

DISCUSSION

Even though RRBs have strong diagnostic significance

due to the clinical complexity of ASD, a better understanding of their characteristics is needed. This study aimed to achieve two objectives. First, we explored the different patterns of RRBs. Through caregiver reports from ADI-R, we discovered that toddlers and young children with ASD exhibited sensory-related RRBs more frequently than other types of RRBs. These findings are consistent with previous studies that have reported a strong association between abnormal sensory responses and RRBs in children with ASD, irrespective of IQ levels or chronological age [28]. Leekham et al. [3] further mentioned that higher-order RRBs (e.g., insistence on sameness) are more likely to develop and be observed in later childhood. Therefore, we may have found lower-order RRBs to be the most common among our participants, all of whom were younger than 42 months.

The second objective was to create subgroups based on RRB coding and investigate their characteristics with other clinical information. Cluster analysis resulted in four groups. Cluster 1 ('multiple RRBs') included non-verbal children with sensory and object interests. According to past studies, these RRBs have been categorized as lower-order RRBs and are associated with general developmental levels and intellectual abilities [3,6]. Therefore, as hypothesized, this group not only scored the highest on instruments for measuring autistic symptom severity but also had the lowest VABS scores across all domains. Similarly, children in Cluster 3 ('sensory RRBs') presented lower-order RRBs but had relatively milder symptoms than those of cluster 1, suggesting that the presence of multiple RRBs may be associated with more developmental delays [9]. Cluster 2 ('resistance to change RRBs'), the largest and oldest group, was comprised of children with resistance to change. Finally, Cluster 4 ('least RRBs') was the smallest group, in which none of the children were reported to have any resistance to change or sensory-related RRBs. This group scored in the 'moderately low' group for all adaptive func-

Table 2. Grouping of participants based on the two-step cluster analysis

	Cluster			
	1 (n=87)	2 (n=102)	3 (n=73)	4 (n=51)
Sex				
Male	68	83	55	43
Female	19	19	18	8
Age (yr)	32.15±6.5	35.76±5.3	33.0±7.0	31.29±7.8
Object interests				
Absent	0	50 (49)	51 (70)	30 (59)
Present	87 (100)	52 (51)	22 (30)	21 (41)
Resistance to change				
Absent	42 (48)	0	73 (100)	51 (100)
Present	45 (52)	102 (100)	0	0
Restricted interest				
Absent	43 (49)	24 (24)	24 (33)	31 (62)
Present	44 (51)	78 (76)	49 (67)	20 (38)
Sensory interests				
Absent	0	37 (36)	0	51 (100)
Present	87 (100)	65 (64)	73 (100)	0
Body movements				
Absent	18 (21)	72 (71)	54 (74)	35 (69)
Present	69 (79)	30 (29)	19 (26)	16 (31)
Verbal				
Absent	0	8 (8)	5 (7)	4 (8)
Present	0	30 (29)	13 (18)	9 (18)
Non-verbal	87 (100)	64 (63)	55 (75)	38 (75)

Data reported as mean±standard deviation or n (%). Cluster 1 (multiple RRBs), Cluster 2 (resistance to change RRBs), Cluster 3 (sensory RRBs), Cluster 4 (least RRBs)

Table 3. Clinical characteristics by cluster

	Cluster*				ANCOVA	
	1	2	3	4	F	Sig
ADOS total[†]	6.95±1.47	6.22±1.70	6.55±1.63	6.27±1.76	2.32	0.076
SA	7.67±1.57	7.04±1.92	7.37±1.88	7.12±1.7	0.99	0.398
RRB	5.80±2.09	4.97±2.42	5.00±2.46	4.71±2.34	3.12	0.026
SRS	68.60±10.63	62.68±9.83	63.94±11.80	58.16±9.12	5.12	0.002
SCQ (lifetime)	18.75±5.15	15.04±6.20	15.41±6.11	12.22±5.54	11.69	<0.001
VABS						
Communication	65.79±11.22	75.76±15.95	70.99±12.91	76.54±14.94	3.27	0.022
Daily life	76.88±10.45	84.23±15.15	79.85±10.77	85.58±15.26	4.12	0.007
Socialization	65.92±10.88	71.74±15.07	70.37±11.03	72.84±18.89	3.12	0.026
Motor	78.98±10.73	82.92±14.53	80.17±11.64	83.34±12.66	2.51	0.059
Total	64.92±10.220	72.82±15.16	68.85±10.87	74.14±15.18	4.73	0.003

Data reported as mean±standard deviation. *Cluster 1 (multiple RRBs), Cluster 2 (resistance to change RRBs), Cluster 3 (sensory RRBs), Cluster 4 (least RRBs); [†]All ADOS scores were analyzed the calibrated severity scores for comparison across different modules. ADOS, Autism Diagnostic Observation Schedule; SA, Social Affect; RRB, Restricted and Repetitive Behaviors; SRS, Social Responsiveness Scale; SCQ, Social Communication Questionnaire; VABS, Vineland Adaptive Behavior Scale

tioning domains, and when compared with cluster 2, their VABS scores did not significantly differ. As explained by previous findings, these higher-order RRBs are recognized as more complex and related to executive functioning skills [29]. Hence, consistent with our results, children with higher-order RRBs are more likely to display greater adaptive skills than those with lower-order RRBs.

We found that different types of RRBs can provide valuable information in differentiating between ASD subgroups. This study is novel in its approach to investigate unique patterns of RRBs and their potential to indicate the level of functioning in toddlers and young children with ASD. However, this study had some limitations. First, the study only used caregiver-reported measures to determine the presence and associations of RRBs in young children with ASD. While caregivers can be reliable reporters of their children's behavior, their answers are still considered vulnerable to recall bias and can be dependent on their awareness of ASD-related risk signs. Observational measures focused on measuring RRBs and adaptive functioning could be regarded as ideal for validating caregiver reports and attaining a better understanding of this clinical phenomenon. Second, the cross-sectional design of this study using only toddler-aged children limits our ability to track the developmental trajectories of autistic symptoms and adaptive functioning. Future studies should examine whether RRBs observed during toddler stages could similarly predict adaptive outcomes in later stages of development. Furthermore, it is important to note that the ADI-R items were binarily coded to focus on whether the behavior was present without considering the severity of the RRBs. We urge researchers to consider the severity of RRBs as well as explore if combining them with other RRBs can have an effect on other clinical characteristics. Lastly, several questionnaires such as SELSI were collected mid-way through the enrollment process of the two larger studies, which made it difficult to use as a covariate variable. Reliable instruments to measure language abilities in toddlers and young children are needed to further explore their potential relationship with RRBs.

Taken together, our findings introduce potential implications in the diagnosis and treatment of individuals with ASD. Under the broad category of RRBs in the ASD phenotype, heterogeneity of RRBs has been shown to emerge at a young age, thereby suggesting ways to design interventions that target specific RRB predictors of autistic symptoms and adaptive difficulties. These results support the importance of distinguishing RRB subtypes based on their distinct relationships with various functioning abilities. Ultimately, evaluating the prognostic value of early RRBs is critical to enhance our understanding and identify novel therapeutic strategies for tod-

dlers and young children with ASD.

CONCLUSION

This study explored the emerging differences in RRB patterns among toddlers and young children with ASD and evaluated their associations with adaptive functioning skills. These results suggest that clinicians and researchers should understand the role of RRBs when building and executing early interventions that target developmental delays. Its potential to determine developmental trajectories can transform current diagnostic and therapeutic approaches for individuals on the spectrum.

Availability of Data and Material

The datasets generated or analyzed during the study are not publicly available as the IRB approved the data to be used within the research team but could be available from the corresponding author on reasonable request.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

Author Contributions

Conceptualization: Hee Jeong Yoo. Data curation: Da-Yea Song, Dabin Kim, Guiyoung Bong. Formal analysis: Da-Yea Song, Dabin Kim. Funding acquisition: Hee Jeong Yoo. Investigation: Da-Yea Song, Hannah Lee, Dabin Kim. Methodology: Da-Yea Song, Hannah Lee, Dabin Kim. Project administration: Da-Yea Song, Dabin Kim, Guiyoung Bong. Supervision: Hee Jeong Yoo. Writing—original draft: Da-Yea Song, Hannah Lee, Dabin Kim. Writing—review & editing: Guiyoung Bong, Jae Hyun Han, Hee Jeong Yoo.

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