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Differences in Language Ability and Emotional-Behavioral Problems according to Symptom Severity in Children with Autism Spectrum Disorder

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Purpose: The aim of this study was to investigate differences in language ability and emotional-behavioral problems according to the severity of social communication impairments (SCI) and restricted and repetitive behaviors (RRB) in children with autism spectrum disorders (ASD).
Materials and Methods: We grouped 113 children with ASD aged 3–12 years according to the severity of SCI and RRB, and investigated language differences and emotional-behavioral problems among the severity groups. If differences in language abilities between the groups were observed, they were further subdivided to examine possible predictors of both receptive and expressive language abilities.
Results: In cluster analyses using subdomains of the Autism Diagnostic Interview-revised, severe SCI individuals showed lower language ability than their milder counterparts, while RRB showed no differences. Receptive and expressive language in the severe SCI group was negatively predicted by social communication and social motivation, respectively. The severe RRB group showed significantly higher levels of anxiety/distress, somatic complaints, thought problems, attention problems, and aggressive behavior, while the severe SCI group was reported to be more withdrawn.
Conclusion: The results of this study suggest that the severity of SCI greatly affects language ability. In children with severe SCI, social communication and social motivation negatively predicted receptive language and expressive language, respectively. Children with severe RRB may have more emotional-behavioral problems that require active intervention.

Key Words: Autism spectrum disorder, symptom severity, language, problem behavior

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INTRODUCTION

Language ability in children with autism spectrum disorder

Autism spectrum disorder (ASD) is a neurodevelopmental disorder defined by social communication impairment (SCI) and the presence of restricted and repetitive behaviors (RRB).[1] In revision of the Diagnostic and Statistical Manual of Mental Disorders (DSM)-IV to DSM-5, a delay in the acquisition and development of verbal language was deliberately eliminated from the diagnostic criteria because research has shown that this characteristic is neither general nor specific to individuals with

ASD.[2] Therefore, it was concluded that a delay in language development or the presence of other language impairments ought to be noted separately under "specifiers" rather than included as criteria for ASD.[3]

However, despite these changes, language delay is one of the first issues noticed by parents of children with ASD, and approximately 63% of children with ASD present with co-occurring language impairment.[4] There is a general consensus that language abilities play an important role in predicting long-term prognosis with respect to adaptive functioning, psychosocial adjustment, peer relationships, and mental health.[5] Magiati and colleagues[6] conducted a systematic review of longitudinal studies examining the outcomes of children with ASD once they reached adulthood. In five studies, better childhood languages skills predicted fewer adult autism symptoms, along with better adaptive functioning and social outcomes. Children with ASD who had higher language abilities were observed to have rates of development similar to those of typically developing children.[5] However, in the same study, children with lower language abilities made slower progress and had flatter trajectories in terms of development. The above reports suggest that ongoing attention and intervention are necessary to target and improve language ability, even though it is excluded from current diagnostic criteria.

Core symptom severity of ASD

The severity of both SCI and RRB also needs to be considered in ASD prognosis. One study observed that the symptom severity of ASD remained stable over time in most children.[7] Accordingly, the authors advised that accurately characterizing and measuring the severity of symptoms at the time of diagnosis, as well as reevaluating symptoms, are essential as they can provide considerable information about the course and prognosis of the disorder. In DSM-5,[1] the meaning of "severity" has been revised to mean "disability in context" rather than "disability in person," indicating that interventions to improve adaptive function are valuable. This new severity rating for SCI and RRB has been found to be associated with behavioral observations of autistic symptomology.[8] Therefore, the Autism Diagnostic Observation Schedule-2 (ADOS-2)[9] and Autism Diagnostic Interview-revised (ADI-R),[10] which are instruments used to measure observed core symptoms in ASD, could be reliable means of rating severity. Indeed, there have been discussions as to whether there are distinct subtypes according to the quality of severity or a gradient according to the quantity or amount of severity.[11] Cluster analysis, which is a statistical method for homogenous grouping based on observed phenotypic similarities and differences, has been applied to ASD symptoms; however, the results thereof have indicated different cluster solutions.[12,13] Despite these discrepancies, rating SCI and RRB dimensions would be clinically meaningful, and homogeneous grouping based on severity within each of the two dimensions would aid in stratification.

Although it has been suggested that the severity of impairments can vary across domains (e.g., core ASD symptoms, cognitive function, adaptive behavior, language ability) in children with ASD,[14] it can be very difficult to separate core symptomology from cognitive function and language ability. Since most observational measures for symptom severity in ASD are strongly influenced by language ability, very little has been reported regarding how the severity of SCI and RRB may be related to language ability.[15] In this context, determining these relationships is a valuable task as it could aid in improving prognosis predictions by increasing the availability of useful information and reducing complexity. However, cross-sectional studies that have examined how symptom severity relates to language ability are limited, at least to the best of our knowledge. If there is a difference in language ability according to core symptom severity, screening individuals who are at higher risk of language delay could be done while individuals are being screened for ASD and during follow-up evaluations. Dividing language into receptive and expressive language and exploring factors that predict each language ability will enable more targeted and optimized interventions for improving language abilities.[16]

Emotional-behavioral problems in children with ASD

The behavioral problems that accompany ASD are diverse, and it is very important to predict who is likely to have maladaptive behaviors, as those with ongoing problems in this area are at a higher risk of long-term mental health issues, such as poor peer relationships and academic achievement.[17] Particularly during childhood and adolescent development, problem behavior often overlaps or coexists with other behaviors, rather than presenting as a single symptom or issue. As it can be more difficult to identify the cause of problem behavior and to predict future developmental differences, internalization-externalization classifications are considered useful. Therefore, regarding the relationship between general ASD symptomology and the Child Behavior Checklist (CBCL), it was reported that the withdrawal subdomain was the most effective for distinguishing young children with ASD (age 36–71 months) from other children.[18] This was supported by another study that found that the total symptom severity of ASD was correlated with problematic behavior.[19] However, few studies have explored which emotional-behavioral problems are associated with the two core symptoms of ASD.

Classification of subgroups according to the severity of symptom domains

The ADI-R is an ideal measure for an autism phenotype study, as it is a comprehensive instrument that includes items for age of onset, language development, SCI, and RRB,[20] and previous studies have conducted cluster analyses on the ADI-R for clinically meaningful stratification. One study showed four phenotypic clusters with multiple clustering methods via 123 item scores from the ADI-R in 1954 individuals with ASD.[12] In contrast, a recent study[13] conducted a cluster analysis for 463 patients with ASD, and three cluster solutions were found to correspond with the severity of SCI and RRB. While the results of the cluster analyses using all items from the ADI-R were not consistent due to confounding factors, such as age, sex, and IQ, cluster analysis has the advantage of creating phenotypically homogenous groupings. In this context, grouping individuals according to the severity of symptoms by performing cluster analyses on each subdomain of the ADI-R that corresponded to SCI and RRB would be meaningful. The ADI-R is divided into three subdomains: the first two, (A) impairments in reciprocal social interaction and (B) impairments in communication, correspond to SCI in DSM-5, and the third, (C) repetitive behaviors and stereotyped pattern, corresponds to RRB in DSM-5.

The current study

The purpose of this study was to investigate differences in language ability and emotional-behavioral problems according to the severity of SCI and RRB in children with ASD. The children were grouped according to the severity of the two core symptom dimensions of ASD, and language differences and emotional-behavioral problems between the groups were investigated. If differences in language abilities between the groups were observed, they were further subdivided to identify possible predictors of both receptive and expressive language abilities.

MATERIALS AND METHODS

Participants

The participants included 113 individuals between the ages of 29 and 144 months who visited the developmental disorder clinic of Child and Adolescent Psychiatry in Severance Hospital, Yonsei University College of Medicine in Seoul, Korea from May 2017 to August 2019. All participants were diagnosed by a child and adolescent psychiatrist using DSM-5 criteria, and all children also met the clinical cutoffs for ASD diagnosis according to ADI-R and ADOS. The participants in this study were Korean, and all family members were proficient native Korean speakers. Demographic and clinical information regarding age, birth history, parental age at birth, symptom recognition point, psychotropic medication, mental health diagnosis, and other medical diagnoses was collected. The Institutional Review Board of the hospital approved the study protocol (approval number: 2019-3350-001).

Measures

Diagnosis of autism spectrum disorder

The ADI-R is a structured interview administered to the parent or primary caregiver to assess the occurrence and patterns of ASD symptoms in early childhood in the domains of communication, social interaction, and restricted interests and repetitive behaviors.[10] One benefit of this measure over other observational measures is that it provides a historical overview of an individual's symptoms in early development as opposed to only observing current symptoms and behavior. The ADI-R consists of three primary subdomains: the first addresses social interaction (A1-A4) and includes questions about nonverbal behavior, peer relationships, shared enjoyment, and reciprocity. The second subdomain, communication (B1-B4), assesses delays in language development or failure to compensate through gestures, lack of social imitative play, difficulties in maintaining conversation, and stereotyped speech. The third subdomain, repetitive and stereotyped behavior (C1-C4), assesses circumscribed interests, nonfunctional routines or rituals, motor mannerisms, and preoccupations with particular parts of objects. In addition to these three subdomains, a score is calculated for abnormality of development evident at/before 36 months (D), which is assessed via five items. In this study, for children over 4 years of age, data from the ever/4–5 diagnostic algorithm were used, and the current/ever algorithm was used for all other children who were younger than 4 years of age. We considered ADI-R results to be suggestive of a diagnosis of ASD if scores were above cut-off values in all domains: 10 for subdomain A, 8 for subdomain B, 3 for subdomain C, and 1 for subdomain D.

The ADOS-2 is a semi-structured, play-based, standardized observational measure in which a clinician rates core impairments in social, communication, and restricted/repetitive behaviors and interests that are associated with ASD.[9] The ADOS-2 is implemented by selecting one of four modules based on the child's chronological age and level of expressive language; therefore, it can be applied to individuals of varying developmental ages and language abilities. The ADOS-2 is divided into two subdomains, social affect (SA) and RRB, and a score is provided for each. The overall total score, combined with SA and RRB scores, was used as the cut-off in each module: 16 in module 1, 10 in module 2, and 9 in module 3.

Symptom-specific measure

The Social Responsiveness Scale (SRS) is a rating scale for individuals between the ages of 4 and 18 years that is completed by a parent or teacher, focusing on the child's behavior in the previous 6 months. Aiming to identify a wide spectrum of deficits in reciprocal social behavior,[21] the SRS is administered as a 65-item questionnaire and can be applied irrespective of the chronological age or language level of the child. Questions pertaining to five subscales are included: social awareness, social cognition, social communication, social motivation, and autistic mannerisms.

It is suggested that this measure can be used for the screening of a child who is at risk for ASD as a way to delineate social characteristics, to generate a behavioral treatment goal, or to target a point of intervention.[22] The SRS generates a total score (SRS-Raw) as well as a sex-normed T-score, which is intended

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to correct for sex differences observed in a normative sample. While both of the scores serve as an index of severity of social deficits, the manual recommends using SRS-Raw in research settings so that comparisons can be made across studies; accordingly, many researchers are following this guide.[23] In the current study, a Korean version of the SRS, with known reliability and validity similar to the original U.S. version, was administered. Raw scores of 195 points were used.

Cognitive assessment

To assess cognitive level, the Korean-Wechsler Preschool and Primary Scale of Intelligence-IV (K-WPPSI-IV) and the Korean-Wechsler Intelligence Scale for Children-IV (K-WISC-IV) were used based on the child's age and degree of language development. Each test was standardized to the Korean version.[24,25] The K-WPPSI-IV and K-WISC-IV provide an overall IQ score (full scale IQ; FSIQ) that represents overall intellectual ability; they also provide index or subtest scores on specific cognitive domains. However, it is difficult to clearly distinguish between verbal and nonverbal IQ because several tasks on the two tests are affected by language ability. Accordingly, in this study, the FSIQ was used to represent cognitive function. A total of 100 children were assessed for cognitive level, of whom 56 were administered the K-WPPSI and 44 were administered the K-WISC.

Language ability

The Sequenced Language Scale for Infants (SELSI) is a Korean semi-structured, examiner-rated scale that is used to indirectly grade language abilities in children who have a developmental age of 4 to 35 months.[26] This is the one and only comprehensive assessment tool for communication and language abilities for toddlers under the age of 3 years in Korea. The SELSI consists of questions that evaluate both receptive and expressive language abilities. The test items assess 1) semantic language associated with cognitive ability, 2) syntactic language associated with linguistic knowledge, and 3) pragmatic language associated with social interaction. On the SELSI, the age range of 4–35 months is divided into 14 developmental age stages, and five questions are answered at each stage. The parents respond to the questions at the child's age level, as well as to questions corresponding to the three levels before and three levels after the child's age level, for a total of 35 questions. This is done for both receptive and expressive language abilities. Therefore, a total of 70 questions are answered. A previous study assessed the reliability and validity of this scale[26] and reported a Cronbach's α, which was calculated to verify the reliability of the internal consistency of the questions, higher than 0.98 for both the receptive and expressive language tests. Furthermore, there were no sex differences between individual questions for either test. Lastly, in investigating the retest reliability, the two tests were performed at intervals of 1 week, and correlations were significant for both receptive (0.996) and expressive language (0.998).

The Preschool Receptive-expressive Language Scale (PRES) is a tool that directly measures the receptive and expressive language of preschool children aged 2–6 years.[27] The test includes items on semantics, syntax, and pragmatics, and uses various test methods involving pictures, objects, instructions, and interviews. To verify the reliability of the scale's internal consistency, Cronbach's α was calculated from the data of 621 children and was found to be 0.95 for both receptive and expressive language. The retest reliability was 0.78 for receptive language and 0.92 for expressive language.[27]

The Receptive and Expressive Vocabulary Test (REVT) is a receptive and expressive vocabulary test that covers the entire life-span from the age of 2 years to adulthood. It is divided into REVT-R (REVT-Receptive), which measures receptive vocabulary, and REVT-E (REVT-Expressive), an expressive vocabulary test. REVT-R is performed by having the individual select a picture, among four, that corresponds to the target vocabulary; in REVT-E, the individual is asked to look at the picture and state the target vocabulary. The REVT-R and REVT-E consist of 188 questions each, and the retest reliability was measured as 0.823 for receptive vocabulary and 0.855 for expressive vocabulary.[28]

Comorbid emotional-behavioral problems

The CBCL is a behavioral rating scale completed by a primary caregiver or teacher that measures emotional-behavioral problems and associated adaptive function in children and adolescents in a standardized format.[29] Korean versions of the CBCL, K-CBCL (1.5–5) and K-CBCL (6–18), were standardized in Korea and are currently in use. Standard scores are created based on T-score criteria according to sex and age group, and the scale is suitable for evaluating children and adolescents along the developmental continuum.

Statistical analyses

IBM SPSS Statistics for Windows, version 21 (IBM Corp., Armonk, NY, USA) was used to conduct all analyses. Descriptive statistics, including mean scores, standard deviations (SD), range, and percentages, were calculated for the demographic variables. For language ability, the measured age equivalent of each language scale divided by the chronological age was used. The raw scores of the ADI-R subdomains were tested for internal consistencies using Cronbach's α. Raw scores for subdomains A1-A4, B1-B4, C1-C4, and D were converted into standardized Z-scores (M=0, SD=1) to achieve comparability across subdomains. Two multiple cluster analyses using each subdomain were sequentially conducted. For the first grouping according to RRB, Z-scores of the C subdomains were used. First, a single linkage analysis was performed to identify and remove outliers. Next, agglomerative hierarchical clustering was conducted while using squared Euclidean distance as proximity and Ward's method for fusion. From this process, the number of clusters was confirmed through a dendrogram, and this number was used in the K-means procedure with the goal of optimizing the cluster solution. For missing values, we excluded cases pairwise in the K-means process. In the second grouping according to SCI, the Z-scores of the A and B subdomains were used. The analysis proceeded in the same manner as that for RRB grouping analysis. After grouping into clusters, comparisons of the cluster means, with respect to demographic and clinical characteristics, ASD diagnostic measures, language ability, and emotional-behavioral problems, were achieved by independent sample *t* tests or χ^2-tests. Lastly, linear regression analyses were performed to determine the predictors of language abilities when there were differences in language ability between groups. Separate analyses were conducted for receptive and expressive language abilities. The predictors included chronological age, sex, IQ, and the five subscales of the SRS.

RESULTS

Descriptive characteristics

In this study, 113 participants were diagnosed with ASD using the DSM-5 diagnostic criteria and the ADOS and/or ADI-R. The sample consisted of 95 (84.1%) male and 18 (15.9%) female individuals aged 29–144 months, with a mean age of 71.41 months (SD=22.54). Cognitive assessment results were available for 96 individuals; IQs ranged from 31–120 (M=65.97, SD=18.29). With respect to medication, 42 (37.2%) patients were taking at least one antipsychotic. The parental age at birth was ascertained for 111 participants, with a mean paternal age of 34.24 years (SD=4.62) and a mean maternal age of 31.69 years (SD=3.67).

ADI-R subscale scores were available for all participants, with total subscores for social interaction (M=19.74, SD=5.21), communication (M=15.09, SD=4.01), stereotyped behavior (M=6.59, SD=2.38), and abnormal development (M=4.13, SD=1.04). Analysis of the ADI-R subdomain scores showed varying reliability, Cronbach's α=0.74 for social interaction, α=0.45 for communication, and α=0.35 for stereotyped behaviors. ADOS-2 scores were available for 112 individuals, and each participant had been administered the module that corresponded to their age and expressive language abilities. Raw scores for ADOS-2 module 1 were available for 21 children (SA: M=15.24, SD=0.65; RRB: M=5.05, SD=0.37); raw scores for module 2 were available for 68 children (SA: M=12.93, SD=0.39; RRB: M=4.35, SD=0.18); and raw scores for module 3 were available for 23 children (SA: M=12.35, SD=0.69; RRB: M=4.21, SD=0.37). Scores of the SRS were available for 83 children, with a mean score of 86.31 (SD=28.11). In regards to language assessment, SELSI was performed in 44 children, PRES in 53 children, and REVT in 50 children. Scores on the K-CBCL were available for n=96 children, with mean total T-score of 64.75 (SD=11.73).

Grouping by the severity of RRB

When grouping individuals based on RRB scores, hierarchical clustering analysis, the resulting dendrogram, and inspection of the reduction in the error sum of squares indicated a two-cluster solution. The subsequent adjusted cluster solution showed the following characteristics: cluster 1 comprised 51 children (45.13%), and cluster 2 comprised 62 children (54.87%). Cluster 1 included children who had high z-scores on the C domain of the ADI-R, indicating that this was a severe RRB group; accordingly, cluster 2 was a mild RRB group. The difference in the C domain between the two groups is shown in Table 1, where the severe RRB group scored higher in all four subcategories.

Table 2 reports the demographic and clinical characteristics of the two subgroups resulting from the cluster analysis based on RRB scores. There were no significant differences in age, sex distribution, IQ, symptom recognition point, and rate of taking antipsychotics. With respect to parental age at birth, paternal age was not different, whereas maternal age was significantly lower in the severe RRB group (p=0.024). In SRS, only autistic mannerism was significantly higher in the severe RRB group (p< 0.001). According to these results, the two groups showed similar overall symptom severity and differed only in terms of RRB. Regarding measures of language abilities, receptive and expressive language abilities did not differ between the groups in any of the three language assessments (SELSI, PRES, or REVT). The severe RRB group showed higher scores for several of the K-CBCL categories, including internalizing (p=0.043), anxious/depressed status (p=0.028), somatic complaints (p=0.040), thought problems (p=0.018), attention problems (p=0.032), and aggressive behavior (p=0.015), and showed a significantly higher overall total score (p=0.010).

Grouping by the severity of SCI

When grouping individuals based on SCI, the hierarchical clustering analysis, resulting dendrogram, and inspection of the reduction in the error sum of squares indicated a two-cluster solution. The subsequent adjusted cluster solution showed the following characteristics: cluster 1 comprised 72 (63.72%) children, and cluster 2 comprised 41 (36.28%) children. Cluster 1 included children who had high z-scores for the A and B subscales of the ADI-R, indicating that this was a severe SCI

Table 1. Comparison of ADI-R Scores between Severe and Mild RRB Groups in Cluster Analysis

Repetitive behaviors and stereotyped pattern	Severe RRB (n=51)	Mild RRB (n=62)	*p* value
C1 Circumscribed interest	0.578 (0.968)	-0.476 (0.748)	<0.001
C2 Nonfunctional routines	0.550 (0.937)	-0.453 (0.809)	<0.001
C3 Motor mannerisms	0.386 (0.914)	-0.317 (0.961)	<0.001
C4 Preoccupations with part-objects	0.565 (0.870)	-0.465 (0.854)	<0.001

ADI-R, Autism Diagnostic Interview-revised; RRB, restricted and repetitive behaviors.
Severe RRB group with higher scores for repetitive behavior and stereotyped pattern dimensions, mild RRB group with lower scores for repetitive behavior and stereotyped pattern dimensions. Data are presented as mean (standard deviation).

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Table 2. Group Comparison according to Sample Characteristics, Autism Specific, Language Profile, and Emotional-Behavioral Problems

	Severe RRB (n=51)	Mild RRB (n=62)	χ^2/*p* value	Severe SCI (n=72)	Mild SCI (n=41)	χ^2/*p* value
Demographic characteristics						
Age (months), n=113	n=51 74.29 (23.07)	n=62 69.84 (21.75)	0.294	n=72 72.54 (20.49)	n=41 70.63 (22.55)	0.665
Sex, n=113	n=51	n=62		n=72	n=41	0.063/0.082
Female/male	6/45	12/50	1.204/0.273	11/61	7/34	
Intelligence, n=100	n=46 67.65 (18.83)	n=54 63.90 (17.80)	0.310	n=65 63.18 (17.58)	n=35 70.17 (18.95)	0.068
Medication, n=113	n=51	n=62		n=72	n=41	
Antipsychotics (%)	19 (37.3)	23 (37.1)	0.001/0.986	34 (47.2)	8 (19.5)	8.59/0.003
Parental age at birth, n=111	n=51	n=60		n=70	n=41	
Paternal age at birth	33.88 (4.68)	34.55 (4.58)	0.450	34.77 (4.57)	33.34 (4.62)	0.116
Maternal age at birth	30.84 (3.60)	32.42 (3.60)	0.024	31.91 (3.58)	31.32 (3.83)	0.410
Onset, n=105	n=48	n=57		n=68	n=37	
Symptom recognition point (months)	34.44 (20.83)	31.23 (15.10)	0.363	31.78 (15.02)	31.32 (3.83)	0.481
Autistic specific symptom						
SRS, n=83	n=35	n=48		n=57	n=26	
Total score	92.78 (31.54)	81.46 (24.47)	0.068	94.36 (27.08)	67.82 (21.03)	<0.001
Social awareness	11.94 (4.85)	11.14 (3.89)	0.409	12.49 (4.39)	9.18 (3.12)	0.001
Social cognition	17.03 (6.78)	15.85 (5.55)	0.389	18.02 (5.95)	12.64 (4.64)	<0.001
Social communication	32.17 (11.32)	30.19 (9.98)	0.401	33.96 (9.76)	24.64 (9.13)	<0.001
Social motivation	14.49 (6.29)	12.90 (4.90)	0.199	15.02 (5.48)	10.14 (4.25)	<0.001
Autistic mannerisms	16.40 (6.85)	11.17 (4.99)	<0.001	14.53 (6.54)	10.68 (5.11)	0.008
Language ability (age equivalent/chronological age)						
SELSI, n=44	n=14	n=30		n=28	n=16	
Receptive	0.532 (0.156)	0.470 (0.221)	0.350	0.407 (0.157)	0.635 (0.196)	<0.001
Expressive	0.500 (0.149)	0.406 (0.185)	0.109	0.363 (0.147)	0.586 (0.138)	<0.001
PRES, n=53	n=28	n=25		n=26	n=27	
Receptive	0.729 (0.202)	0.719 (0.189)	0.862	0.676 (0.186)	0.770 (0.193)	0.086
Expressive	0.676 (0.207)	0.663 (0.710)	0.811	0.636 (0.193)	0.703 (0.183)	0.196
REVT, n=50	n=24	n=26		n=27	n=23	
Receptive	0.846 (0.211)	0.804 (0.248)	0.525	0.804 (0.225)	0.847 (0.237)	0.515
Expressive	0.819 (0.291)	0.824 (0.255)	0.936	0.802 (0.253)	0.845 (0.271)	0.561
Emotional-behavioral problem						
K-CBCL T score, n=96	n=41	n=55		n=63	n=33	
Total problem score	68.46 (13.24)	61.98 (9.68)	0.010	66.68 (11.86)	61.06 (10.69)	0.025
Internalizing broad band score	62.76 (12.62)	58.02 (10.01)	0.043	62.32 (11.71)	55.69 (9.45)	0.006
Externalizing broad band score	64.83 (12.45)	60.89 (11.07)	0.105	63.25 (12.10)	61.27 (11.22)	0.437
Attention problems	69.32 (11.03)	65.09 (7.95)	0.032	68.24 (10.08)	64.33 (8.04)	0.057
Aggressive behavior	64.24 (10.70)	59.20 (9.25)	0.015	61.78 (10.39)	60.55 (9.80)	0.575
Anxious/depressed	60.54 (9.92)	56.33 (7.89)	0.028	59.41 (9.34)	55.67 (7.91)	0.053
Somatic complaints	56.36 (6.97)	53.62 (5.48)	0.040	55.21 (6.37)	54.00 (6.11)	0.374
Withdrawn	65.49 (10.98)	65.02 (8.91)	0.818	67.75 (10.32)	60.39 (6.48)	<0.001
K-CBCL (1.5–5 yr), n=53	n=21	n=32		n=33	n=20	
Emotionally reactivity	60.14 (7.97)	55.34 (6.12)	0.017	58.58 (7.32)	55.05 (6.70)	0.086
Sleep problems	58.57 (10.32)	56.81 (7.71)	0.481	58.15 (9.49)	56.45 (7.61)	0.500
K-CBCL (6–18 yr), n=43	n=20	n=23		n=30	n=13	
Social problems	70.75 (10.27)	67.83 (7.30)	0.283	70.73 (8.13)	65.62 (9.63)	0.080
Thought problems	67.50 (9.69)	60.57 (8.77)	0.018	64.33 (10.36)	62.54 (8.41)	0.585
Rule-breaking behavior	60.45 (8.83)	60.09 (5.89)	0.877	60.80 (7.92)	59.00 (5.76)	0.465

RRB, restricted and repetitive behaviors; SCI, social communication impairments; SRS, Social Responsiveness Scale; SELSI, Sequenced Language Scale for Infants; PRES, preschool receptive-expressive language scale; REVT, Receptive and Expressive Vocabulary Test; K-CBCL, Korean Child Behavior Checklist.
Data are presented as mean (standard deviation).

group, while cluster 2 was a mild SCI group. The differences in the scores for A and B domains between the two groups are shown in Table 3, whereas the severe SCI group scored significantly higher in all areas of the A and B subscales of ADI-R, except for B3 (stereotyped speech).

Age, sex, IQ, parental age at birth, and symptom recognition points were not significantly different between the groups; however, the rate of taking antipsychotics was higher in the severe SCI group (χ^2=8.59, p=0.003) (Table 2). In terms of language abilities, both the REVT and PRES showed lower language ability in the severe SC group, although this difference was not significant. On the SELSI, both receptive and expressive language abilities were significantly lower in the severe SCI group (p<0.001, p<0.001). Finally, on the K-CBCL, the total problem score (p=0.025) and scores for internalizing (p=0.006) and withdrawal (p<0.001) were higher in the severe SCI group; there were no differences in the scores for any other areas of the K-CBCL.

Since language ability differed according to the severity of SCI symptoms, stepwise multiple regression analyses were conducted to assess concurrent predictors of receptive and expressive language abilities in the severe SCI group. Eleven predictor variables were entered into the model: age, sex, IQ, use of antipsychotics, and the five subscores of the SRS. Receptive and expressive language abilities were used as dependent variables. As seen in Table 4, a one-step regression model significantly predicted receptive language ability in the severe SCI group. On the SRS, social communication accounted for 22.1% of the variance in the severe SCI group's composite receptive language ability. A two-step model significantly predicted expressive language ability in the severe SCI group. Two predictors, IQ and social motivation in the SRS, accounted for 51.4% of the variance in the severe SC group's composite expressive language ability.

DISCUSSION

In this cross-sectional study, we explored how the severity of SCI and RRB, the core symptoms of ASD, are related to language ability and emotional-behavioral problems. In the present study, we noted no differences in language ability according to the severity of RRB; however, language ability was significantly lower in children with severe SCI. When examining the factors that predicted language proficiency in the severe SCI group, we found that social communication influenced receptive language and that social motivation and IQ influenced expressive language. Secondly, in terms of emotional-behavior problems, the severe RRB group scored significantly higher in anxiety/distress, somatic complaints, thought problems, attention problems, and aggressive behavior than the mild RRB group, while the severe SCI group was more withdrawn than the mild

Table 3. Comparison of ADI-R Scores between Severe and Mild SCI Groups in Cluster Analysis

	Severe SCI (n=72)	Mild SCI (n=41)	*p* value
Impairments in reciprocal social interaction			
A1 Nonverbal behaviors	0.481 (0.784)	-0.844 (0.752)	<0.001
A2 Peer relationships	0.369 (0.818)	-0.648 (0.968)	<0.001
A3 Shared enjoyment	0.532 (0.510)	-0.935 (0.965)	<0.001
A4 Reciprocity	0.400 (0.757)	-0.703 (0.992)	<0.001
Impairments in communication			
B1 Delay of language	0.461 (0.793)	-0.810 (0.792)	<0.001
B2 Conversation	0.368 (0.673)	-0.517 (1.15)	<0.001
B3 Stereotyped speech	0.087 (1.06)	-0.122 (0.902)	0.336
B4 Imitative play	0.541 (0.581)	-0.951 (0.864)	<0.001

ADI-R, Autism Diagnostic Interview-revised; SCI, social communication impairments.
Severe SCI group with higher scores for impairment in social and communication dimensions, mild SCI group with lower scores for impairment in social and communication dimensions. Data are presented as mean (standard deviation).

Table 4. Linear Regression Analysis for Predictors of Receptive and Expressive Language Ability in the Severe SCI Group

Risk factor	B (SE)	β	F	*p* value	R^2	ΔR^2
Receptive language ability in SELSI						
1 SRS–social communication	-0.008 (0.003)	-0.516	5.821	0.028	0.221	0.140
Expressive language ability in SELSI						
1 IQ	0.008 (0.002)	0.637	10.898	0.005	0.368	0.108
2 IQ	0.008 (0.002)	0.610	9.986	0.003		
SRS–social motivation	-0.013 (0.005)	-0.408		0.029	0.514	0.094

SCI, social communication impairments; SELSI, Sequenced Language Scale for Infants; SRS, Social Responsiveness Scale.
Model was adjusted for age, sex, IQ, use of antipsychotics, and the five subscores of the SRS.

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SCI group.

Previous research, notably longitudinal studies, has described a relationship between RRB symptom severity and language.[30,31] However, these studies have shown inconsistent results depending on the age of the participants. Paul, et al.[30] followed 37 toddlers diagnosed with ASD for 2 years and found that RRBs using the ADOS at 22 months significantly predicted expressive language at 47 months. This suggests that toddlers with prominent RRBs are at higher risk for language impairments and require early intervention. In a study of 115 children with ASD controlling for gains in nonverbal cognitive function, Ray-Subramanian and Ellis Weismer[31] showed that increased receptive and expressive language skills from the ages of 2 to 3 years predicted a decrease in RRB at age 3. Taken together, these results suggest that young children who are able to acquire language skills with which to be able to better understand and communicate with others in their environment may require fewer RRBs as a means of self-soothing.

However, as alluded to above, the results were different in studies involving older children with ASD. In a longitudinal study, preschool children were assessed initially on the severity of social affect and RRB as measured by the ADOS, as well as on language ability, and assessed again 1 year later. Therein, social affect-calibrated severity score was a significant factor in predicting expressive language a year later, while RRB severity score was not.[32] One retrospective study employed data from a of minimally verbal children who were at least 8 years old (M=11.6 years, SD=2.73 years) and who had not acquired phrase speech until 4 years of age. Logistic regression analyses were performed to explore the predictors of phrase/fluent speech acquisition, and unlike social impairment, RRB as measured by the ADI-R appeared to be not significantly associated with delayed speech attainment.[33] Altogether, this suggests that for younger children with ASD, severity of RRB affects language development and that language acquisition is associated with improvements in RRB. However, in older children, the severity of RRB does not appear to have a significant effect on developing fluent speech.

In this regard, we attempted to investigate whether RRB and language ability have a significant relationship at one point in time, rather than examining a change in language abilities and severity of RRB over time. As explained above, since the effect of the severity of RRB on language ability may vary by age, differences in language ability according to severity of RRB would not have been observed [average age of language measure (months)-SELSI: M=49.43, SD=14.03; PRES: M=56.24, SD=15.11; REVT: M=62.54, SD=20.16]. However, the rate of SELSI in the mild RRB group was 48.4%, whereas it was only 27.5% in the severe RRB group, and this may have affected the results.

In terms of SCI, our results may be more meaningful considering that the language ability, measured by SELSI, was lower in the group with higher SCI severity, and there was no significant difference in IQ between the groups. A previous cross-sectional study[33] of children with ASD over the age of 8 years who did not speak in phrases until the age of 4 years demonstrated a similar relationship between social impairment and language ability. In that study, social impairment, as assessed using the ADI-R, influenced the age at which the child started to speak in phrases and was also a factor in predicting fluent speech.[33] According to the longitudinal study by Thurm, et al.[32] mentioned earlier, improvement in the social affect-calibrated severity score of the ADOS was predictive of expressive language by the age of 5 years, whereas the RRB-calibrated severity score was not related to language outcomes. In summary, previous studies have found that social affect has more of an effect on the development of language abilities than RRB, a finding which we corroborate here. This study also demonstrated that language ability was lower in children with severe SCI, even when viewed cross-sectionally across several ages. These results suggest that children with severe SCI may be at a higher risk of language delay, for which active language therapy may be required.

Overall language abilities, as measured on the PRES and REVT, were lower in the severe SCI group, although this difference was not significant. The PRES and REVT are heavily influenced by cognitive functioning as the instructions can be difficult for children to understand, while the SELSI is based on parental reporting. Therefore, children examined by the PRES or REVT would have relatively intact cognitive functioning, such that the difference in language ability according to symptom severity may not be noticeable. In this context, the SELSI is a more useful tool for measuring actual language skills without the influence of cognitive function, and differences therein may be more clinically meaningful.

In multiple regression analyses for children in the severe SCI group, we found that social communication scores in the SRS negatively predicted receptive language ability. That is, social communication was related to receptive language, not social awareness, representing the ability to pick up a social cue or social cognition indicating the extent to which the social cue was interpreted. Social communication represents the expressive aspects of reciprocal social behavior.[22] It includes the following items: “awkward in turn-taking interaction with peers,” “has trouble keeping up with the flow of a normal conversation,” “too silly or laughs inappropriately,” “talks to people with an unusual tone of voice,” “emotionally distant,” and so on.[22] Therefore, the motoric aspect of communication may not only be related to expressive language but also deeply related to receptive language in terms of reciprocity. This can also be understood in this context that in children with ASD, expressive language is more dominant than receptive language, unlike in typical development or language delay.[34] Accordingly, the greater the SCI in severe SCI group, the greater the receptive language delay, and thus, the relationship with the expressive language may be reversed.

Similar to previous studies, expressive language was found to be affected by IQ and negatively predicted by social motivation score on the SRS. Thus, children with deficits in social motivation may have low expressive language abilities. Social motivation, as defined on the SRS, is a measure of how motivated a child may be to interact socially with others and includes constructs of social anxiety, inhibition, and empathic orientation.[22] The items on this subscale contain the following: "would rather be alone than with others," "does not join group activities unless told to do so," "too tense in social settings," "avoids people who want to be emotionally close to him," "clings to adults, seems too dependent on them," and so on.

There are a few studies that have examined the relationship between anxiety and language, and most of them have focused on how language ability affects anxiety. Davis, et al.[35] showed that receptive language was positively associated with anxiety scores in young children with ASD; they interpreted that the higher the language ability, the better the understanding of negative social information, which would then be translated into anxiety. Other studies have focused on pragmatic language rather than structural language. Boonen, et al.[36] hypothesized that a deficit in pragmatic language leads an individual to misperceive social situations; thus, the child feels insecure and has increased anxiety, accompanied by internalizing behaviors. Similarly, a study that included 159 children with ASD aged between 4 and 7 years reported that while structural language positively predicted anxiety level, pragmatic language was inversely related to anxiety and externalizing behaviors.[37] In addition, the authors found that the more structural language preceded pragmatic language, the more prone children were to exhibit anxiety symptoms.

Unlike previous studies that predicted a child's anxiety level based on their language ability, in this study, social motivation was found to predict expressive language ability. Anxiety is at the core of social motivation, which in turn can be assumed to affect expressive language. In clinical settings, there are many cases of children with high anxiety, whose receptive language is relatively intact while expressive language development is delayed, although few studies have investigated this. This study reflects what has been observed in clinical settings and highlights that both receptive language and expressive language are closely related to anxiety.

In this study, children in the severe RRB group scored high for anxiety/distress, somatic complaints, thought problems, attention problems, and aggressive behavior, and consequently, both internalizing and externalizing behaviors were prominent. This suggests that children with severe RRB are likely to also have challenging behaviors and may require active intervention. In particular, when investigating which of these behavioral issues were significantly related to antipsychotic prescriptions in all participants, aggressive behavior was found to have the highest association. When considering SCI, the severe SCI group was significantly more socially withdrawn than the than mild SCI group. In summary, compared to SCI, the severity of RRB appears to be associated with more diverse emotional behavior problems, which would affect prognosis of the child developing and maintaining peer relationships. Being able to predict who on the spectrum is more likely to have maladaptive behavior based on symptom severity is important from the perspective of delivering appropriate interventions.

It should be noted that this is the first study to investigate how symptom severity in children with ASD relates to language ability and emotional-behavioral problems using a cross-sectional design. Notably, the same specialists administered the diagnostic tests, such as the ADOS-2 and the ADI-R, as well as the language assessments, which make the results more reliable. Also, the clinical features associated with ASD were reviewed in parallel simultaneously, which can be of great help in better understanding patients and setting treatment goals in the clinical setting.

There are a few limitations to this study that should be acknowledged. First, the ADI-R score may have been affected by recall errors as it relies on parental reports. As the raw scores of the ADI-R were not normalized for the purpose of stratification or grouping based on core symptoms, the scores were converted to Z-scores for analysis in this study. However, the major limitation is that the Cronbach's α values of each subdomain of the ADI-R were lower than the clinically significant level. Second, ages ranged from 29 to 144 months and were analyzed together, and some participants were unable to perform certain measures due to age criteria. Despite an attempt to make a common interpretation across a wide age range, there is a limitation to analysis by developmental age. Another possible limitation is that the number of children using the same language measurement was relatively small due to different measures needing to be used based on the child's language level. Nevertheless, although the SELSI is used for children aged 4–35 months, it was a suitable instrument for this study because the children in this study, while having a wider range of ages from 29 to 144 months, had language abilities that were equivalent to 4–35 month old typically developing children. Lastly, 37% of the participants were taking antipsychotics that may affect emotional-behavioral problems, and interventions like language therapy that could affect outcome were not controlled. Other factors, such as comorbidity and parental factors, were not included, and it is difficult to reveal a causal relationship due to the characteristics of a cross-sectional study. Future studies should confirm how symptom severity correlates with language ability and emotional-behavioral problems when controlled with medication and specialized therapy at a certain developmental age.

In conclusion, this study suggests that the severity of SCI greatly affects language ability. In children with severe SCI, social communication and social motivation negatively predicted receptive language and expressive language, respectively. Children with severe RRB may have various emotional-behavioral problems, requiring active intervention.

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