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thought problems, attention problems, and aggressive behavior, while the severe SCI group was reported to be more withdrawn.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
ASD. 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.

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. 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. Magiati and colleagues conducted a systematic review of longitudinal studies examining the outcomes of children with ASD once they reached adulthood. In five studies, better childhood language 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. 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. 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, 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 symptomatology. Therefore, the Autism Diagnostic Observation Schedule-2 (ADOS-2) and Autism Diagnostic Interview-revised (ADI-R), 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. 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. Despite these discrepancies, rating SCI and RRB dimensions would be clinically meaningful, and homogenous 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, 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. 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.

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. 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. This was supported by another study that found that the total symptom severity of ASD was correlated with problematic behavior. 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, 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. In contrast, 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.
Language and Symptom Severity in ASD

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. 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. 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, 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. The SRS generates a total score (SRS-Raw) as well as a sex-normed T-score, which is intended...
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 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 scale27 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
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 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 REVLT). 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 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 REVLT). 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 RRB
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| 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.
Table 2. Group Comparison according to Sample Characteristics, Autism Specific, Language Profile, and Emotional-Behavioral Problems

| Demographic characteristics | Severe RRB (n=51) | Mild RRB (n=62) | χ²/p value | Severe SCI (n=72) | Mild SCI (n=41) | χ²/p value |
|-----------------------------|------------------|----------------|------------|------------------|----------------|------------|
| Age (months), n=113 | 74.29 (23.07) | 69.84 (21.75) | 0.294 | 72.54 (20.49) | 70.63 (22.55) | 0.665 |
| Sex, n=113 | 0.063/0.082 | 0.063/0.082 | 1.204/0.273 | 11/61 | 7/34 |
| Intelligence, n=100 | 67.65 (18.83) | 63.30 (17.80) | 0.310 | 63.18 (17.58) | 70.17 (18.95) | 0.068 |
| Medication, n=113 | 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 | 33.88 (4.68) | 34.55 (4.58) | 0.450 | 34.77 (4.57) | 33.34 (4.62) | 0.116 |
| Maternal age at birth | 0.024 | 0.024 | 0.199 | 0.13 | 0.13 |
| Onset, n=105 | 34.44 (20.83) | 31.23 (15.10) | 0.363 | 31.78 (15.02) | 31.32 (3.83) | 0.410 |
| Symptom recognition point (months) | 11/61 | 7/34 | 11/61 | 7/34 | 11/61 | 7/34 |
| Autistic specific symptom | 0.068 | 0.068 | 0.068 | 0.068 | 0.068 | 0.068 |
| SRS, n=83 | 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 | 33.36 (8.76) | 24.64 (9.13) | <0.001 |
| Social motivation | 14.49 (6.23) | 12.90 (4.90) | 0.199 | 15.02 (5.48) | 10.14 (4.25) | <0.001 |
| Autistic mannerisms | 0.401 | 0.401 | 0.401 | 0.401 | 0.401 | 0.401 |
| Language ability (age equivalent/chronological age) | 0.068 | 0.068 | 0.068 | 0.068 | 0.068 | 0.068 |
| SELSI, n=44 | 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 | 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 | 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 | 0.068 | 0.068 | 0.068 | 0.068 | 0.068 | 0.068 |
| K-CBCL T score, n=96 | 68.46 (13.24) | 61.98 (8.68) | 0.010 | 66.68 (11.86) | 61.06 (10.69) | 0.025 |
| Total problem score | 62.76 (12.62) | 58.02 (10.01) | 0.043 | 62.32 (11.71) | 55.69 (9.45) | 0.006 |
| Internalizing broad band score | 64.83 (12.45) | 60.89 (11.07) | 0.105 | 65.25 (12.10) | 61.27 (11.22) | 0.437 |
| Aggressive behavior | 69.32 (11.03) | 65.59 (7.95) | 0.032 | 68.24 (10.08) | 64.33 (8.04) | 0.057 |
| Anxious/depressed | 60.54 (9.92) | 56.33 (7.89) | 0.028 | 59.41 (8.34) | 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 (6.68) | <0.001 |
| K-CBCL (1.5–5 yr), n=53 | 60.14 (7.97) | 55.34 (6.12) | 0.017 | 56.58 (7.32) | 55.05 (6.70) | 0.086 |
| Social problems | 58.57 (10.32) | 56.81 (7.71) | 0.481 | 58.15 (9.49) | 56.45 (7.81) | 0.500 |
| 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 ($\chi^2=8.59, p=0.003$) (Table 2). In terms of language abilities, both the REVTr 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 RRB severity; 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-behavioral 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

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

| Impairments in reciprocal social interaction | Severe SCI (n=72) | Mild SCI (n=41) | $p$ value |
|---------------------------------------------|------------------|----------------|-----------|
| 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)     | $\beta$ | F     | $p$ value | $R^2$ | $\Delta R^2$ |
|------------------------------------|------------|---------|-------|-----------|-------|--------------|
| Receptive language ability in SELSI| -0.008 (0.003) | -0.516  | 5.821 | 0.028     | 0.221 | 0.140        |
| Expressive language ability in SELSI| 0.008 (0.002) | 0.637   | 10.898| 0.005     | 0.368 | 0.108        |
| SRS–social communication           | 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.
SCI group.

Previous research, notably longitudinal studies, has described a relationship between RRB symptom severity and language. However, these studies have shown inconsistent results depending on the age of the participants. Paul, et al. 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 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. 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. 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-sec-
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. 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. 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. 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. 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 predict receptive language and expressive language, respectively. Children with severe RRB may have various emotional-behavioral problems, requiring active intervention.
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REFERENCES

1. American Psychiatric Association. Diagnostic and statistical manual of mental disorders: DSM-5, 5th ed. Arlington (VA): American Psychiatric Association; 2013.
2. Lord C, Bishop SL. Autism spectrum disorders: diagnosis, prevalence, and services for children and families. Social Policy Report 2010;24.
3. Lord C, Bishop SL. Recent advances in autism research as reflected in DSM-5 criteria for autism spectrum disorder. Annu Rev Clin Psychol 2015;11:53-70.
4. Kwok EY, Brown HM, Smyth RE, Cardy JO. Meta-analysis of receptive and expressive language skills in autism spectrum disorder. Res Autism Spectr Disord 2015;9:202-22.
5. Tek S, Mesite L, Feln D, Naigles L. Longitudinal analyses of expressive language development reveal two distinct language profiles among young children with autism spectrum disorders. J Autism Dev Disord 2014;44:75-89.
6. Magiatii I, Tay NW, Howlin P. Cognitive, language, social and behavioural outcomes in adults with autism spectrum disorders: a systematic review of longitudinal follow-up studies in adulthood. Clin Psychol Rev 2014;34:73-86.
7. Gotham K, Pickles A, Lord C. Trajectories of autism severity in children using standardized ADOS scores. Pediatrics 2012;130:e1278-84.
8. Mazurek MO, Lu F, Macklin EA, Handen BL. Factors associated with DSM-5 severity level ratings for autism spectrum disorder. Autism 2019;23:468-76.
9. Lord C, Rutter M, DiLavore P, Risi S, Gotham K, Bishop S. Autism diagnostic observation schedule, 2nd edition (ADOS-2). Torrence (CA): Western Psychological Services; 2012.
10. Rutter M, Le Couteur A, Lord C. Autism diagnostic interview-revised. Los Angeles (CA): Western Psychological Services; 2003.
11. Syropoulos-Delli CK, Papaefstathiou E. Review of cluster analysis of phenotypic data in Autism Spectrum Disorders: distinct subtypes or a severity gradient model? Int J Dev Disabil 2020;66:13-21.
12. Hu VW, Steinberg ME. Novel clustering of items from the Autism Diagnostic Interview-Revised to define phenotypes within autism spectrum disorders. Autism Res 2009;2:67-77.
13. Cholemkery H, Medda J, Lempp T, Freitag CM. Classifying autism spectrum disorders by ADI-R: subtypes or severity gradient? I Autism Dev Disord 2016;46:2327-39.
14. Di Rezze B, Rosenbaum P, Zwaigenbaum L. What attributes determine severity of function in autism? A web-based survey of stakeholders. Focus Autism Other Dev Disabl 2012;27:39-41.
15. Anderson DK, Lord C, Risi S, DiLavore PS, Shulman C, Thurm A, et al. Patterns of growth in verbal abilities among children with autism spectrum disorder. J Consult Clin Psychol 2007;75:594-604.
16. Paul R, Campbell D, Gilbert K, Tsiori I. Comparing spoken language treatments for minimally verbal preschoolers with autism spectrum disorders. J Autism Disord 2013;43:418-31.
17. Lindor E, Sivarathnam C, May T, Stefanac N, Howells K, Rinehart N. Problem behavior in autism spectrum disorder: considering core symptom severity and accompanying sleep disturbance. Front Psychiatry 2019;10:487.
18. Sikora DM, Hall TA, Hartley SL, Gerrard-Morris AE, Cagle S. Does parent report of behavior differ across ADOS-G classifications: analysis of scores from the CBCL and GARS. J Autism Dev Disord 2008;38:440-8.
19. Jang J, Dixon DR, Tarbox C, Granpeesheh D. Symptom severity and challenging behavior in children with ASD. Res Autism Spectr Disord 2011;5:1028-32.
20. Snow AV, Lecavalier L, Houts C. The structure of the Autism Diagnostic Interview-Revised: diagnostic and phenotypic implications. J Child Psychol Psychiatry 2009;50:734-42.
21. Constantino JN. Social Responsiveness Scale. Torrance (CA): Western Psychological Services; 2005.
22. Constantino JN, Gruber CP. Social Responsiveness Scale-second edition (SRS-2). Torrance (CA): Western Psychological Services; 2012.
23. Hus V, Bishop S, Gotham K, Huerta M, Lord C. Factors influencing scores on the social responsiveness scale. J Child Psychol Psychiatry 2013;54:216-24.
24. Kwak K, Oh S, Kim C. Korean-Wechsler Intelligence Scale for Children-IV. Seoul: Hakjisa; 2011.
25. Park H, Lee K, Lee SH, Park M. A study on standardization of K-WPPSI-IV: analyses of reliability and validity. Koreal J of Child Stud 2016;12:111-30.
26. Kim YT. Content and reliability analyses of the Sequenced Language Scale for Infants (SELSI). Commun Sci Discor 2002;7:1-23.
27. Kim YT. Content and reliability analyses of the preschool receptive-expressive language scale (PRE5). Commun Sci Discor 2000;5:1-25.
28. Kim YT, Hong G, Kim K, Jang H, Lee J. Receptive & expressive vocabulary test (REV5). Seoul: Seoul Community Rehabilitation Center; 2000.
29. Achenbach TM, Rescorla LA. Manual for the ASEBA preschool forms and profiles. Burlington (VT): University of Vermont, Research Center for Children, Youth, and Families; 2000.
30. Paul R, Chavarska K, Cicchetti D, Volkmar F. Language outcomes of toddlers with autism spectrum disorders: a two year follow-up. Autism Res 2008;1:97-107.
31. Ray-Subramanian CE, Ellis Weismer S. Receptive and expressive language as predictors of restricted and repetitive behaviors in young children with autism spectrum disorders. J Autism Dev Disord 2012;42:2113-20.
32. Thurm A, Manwaring SS, Swineford L, Farmer C. Longitudinal study of symptom severity and language in minimally verbal children with autism. J Child Psychol Psychiatry 2015;56:97-104.
33. Wodka EL, Mathy P, Kalb L. Predictors of phrase and fluent speech in children with autism and severe language delay. Pediatrics 2013;131:e1128-34.
34. Seol KL, Song SH, Kim KL, Oh ST, Kim YT, Im WY, et al. A comparison of receptive-expressive language profiles between toddlers with autism spectrum disorder and developmental language delay. Yonsei Med J 2014;55:1721-8.
35. Davis TE 3rd, Moree BN, Dempsey T, Hess JA, Jenkins WS, Fodstad JC, et al. The effect of communication deficits on anxiety symptoms in infants and toddlers with autism spectrum disorders. Behav Ther 2012;43:142-52.
36. Boonen H, Maljaars J, Lambrechts G, Zink I, Van Leeuwen K, Nopens I. Behavior problems among school-aged children with autism spectrum disorder: Associations with children’s communication difficulties and parenting behaviors. Res Autism Spectr Disord 2014;8:716-25.
37. Rodas NV, Eisenhower A, Blacher J. Structural and pragmatic language in children with ASD: longitudinal impact on anxiety and externalizing behaviors. J Autism Dev Disord 2017;47:3479-88.