Early Social-Communicative and Cognitive Development of Younger Siblings of Children With Autism Spectrum Disorders

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Objective: To compare the early social-communicative development of younger siblings of children with autism spectrum disorders (ASDs) with that of younger siblings of children with typical development, using parental report and child-based measures.

Design: Group comparison.

Setting: Vanderbilt University, between July 1, 2003, and July 31, 2006.

Participants: Younger siblings of children with ASD (n=64) and younger siblings of children with typical development (n=42) between the ages of 12 and 23 months (mean, 16 months).

Main Exposure: Having a sibling with an ASD.

Outcome Measures: Child-based measures included a cognitive assessment; an interactive screening tool assessing play, imitation, and communication; and a rating of autism symptoms. Parental report measures were an interview of social-communicative interactions and a questionnaire assessing language and communication skills.

Results: Younger siblings of children with ASD demonstrated weaker performance in nonverbal problem solving (mean difference [MD], 5.91; 95% confidence interval [CI], 2.48-9.34), directing attention (MD, 0.52; 95% CI, 0.07-0.97), understanding words (MD, 3.11-63.48), understanding phrases (MD, 4.96; 95% CI, 1.85-7.27), gesture use (MD, 1.49; 95% CI, 0.51-2.47), and social-communicative interactions with parents (MD, 1.32; 95% CI, 0.27-2.37), and had increased autism symptoms (MD, 2.54; 95% CI, 1.05-4.03), relative to control siblings. A substantial minority of the ASD sibling group exhibited lower performance relative to controls. Significant correlations between child-based measures and parental reports assessing similar constructs were found (r=-0.74 to 0.53; P range, .000-.002).

Conclusion: The weaker performance found for children in the ASD sibling group may represent early-emerging features of the broader autism phenotype, thus highlighting the importance of developmental surveillance for younger siblings.

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Sibs-ASD allows for longitudinal investigation of development using direct observations and standard assessments. This approach can provide information not only about the earliest signs of autism but also about the development, manifestation, and boundaries of specific behaviors that may represent the broader autism phenotype, such as social and communication skills.

Recent studies have found that Sibs-ASD demonstrate differences in social and communicative development by the age of 14 to 18 months, such as less advanced levels of requesting, initiating and responding to joint attention, and language and gesture development, relative to siblings of typically developing children (Sibs-TD). Moreover, the latter differences exist even when children who are later diagnosed as having an ASD or language delays are excluded from analyses, suggesting the pervasiveness of early communicative differences in Sibs-ASD. Because children with autism often have learning delays compared with their low-risk peers, cognitive functioning is another area warranting investigation. Information about early performance on standardized cognitive measures is available only for the subgroup of Sibs-ASD who are later diagnosed as having an ASD, or for language subscales only. Thus, the extent to which differences in verbal and nonverbal cognitive abilities exist within the broader group of high-risk siblings, relative to control siblings, is not yet known.

The present study compares the early social-communicative and cognitive development of Sibs-ASD and Sibs-TD. This study extends our knowledge by using a relatively large sample to examine group differences and within-group patterns of performance for child-based and parental report measures.

### METHODS

**PARTICIPANTS**

This study included 106 participants: 64 Sibs-ASD and 42 Sibs-TD. Eligible participants had (1) a chronological age between 12 and 23 months, inclusive; (2) no severe sensory or motor impairments that would impede completion of research assessments; (3) no identified metabolic, genetic, or progressive neurological disorders; (4) English as the primary language; and (5) an older sibling. Eligibility for the Sibs-ASD group required that the older sibling (ie, proband) be diagnosed as having autism, pervasive developmental disorder not otherwise specified, or Asperger disorder; proband diagnoses and available supporting information were obtained from parents. Eligibility for the Sibs-TD group required that the older sibling have no developmental disorders and that there be no family history of autism or mental retardation in first-degree relatives; this information was obtained through a family history interview with parents.

Children were recruited between July 1, 2003, and July 31, 2006. The Sibs-ASD were recruited from university-based autism (n = 34) and speech-language (n = 16) programs, and community agencies (n = 14). The Sibs-TD were recruited through a birth record database (n = 18), university-based research programs (n = 13), and community agencies (n = 11). The research protocol received approval from the Vanderbilt University institutional review board, and all parents signed informed consent forms before beginning any research procedures.

The sample was 87% white and 57% male. Parental occupations ranged from semiskilled to professional, and nearly all mothers (99%) completed high school (Table 1). The groups differed significantly on maternal education (mean difference [MD], 0.42; 95% confidence interval [CI], 0.08-0.76), with mothers of Sibs-TD having a higher mean educational level than mothers of Sibs-ASD. The number of older siblings was comparable in the Sibs-ASD (mean, 1.8) and Sibs-TD (mean, 1.6) groups (MD, 0.23; 95% CI, −0.11 to 0.56). Of the probands, 40 (63%) had autism, 21 (33%) had pervasive developmental disorder not otherwise specified, and 2 (4%) had Asperger syndrome. (Number of probands does not add to 64 because 2 Sibs-ASD came from the same family.)

### PROCEEDURES

All assessments and interviews were conducted or supervised by experienced licensed psychologists, in collaboration with reliably trained graduate students (C.R.M. and others) or research assistants. With the exception of the Childhood Autism Rating Scale (CARS), each measure was administered by a different member of the research team in a single 3-hour session. In rare cases, families were rescheduled for a second session because of child fatigue. Examiners were not blind to sibling group.

### MEASURES

**Mullen Scales of Early Learning**

The Mullen Scales of Early Learning (MSEL) is a measure of cognitive function designed for children from birth through the age of 68 months. Cognitive subscales measure the domains of nonverbal problem solving (visual reception), fine motor skills, receptive language, and expressive language. The MSEL provides a t score (mean, 50; SD, 10) for each domain and an overall early learning composite (mean, 100; SD, 15). The MSEL has strong concurrent validity with other cognitive and language measures and is used commonly with young children with autism. The 4 t scores and the early learning composite were used to measure cognitive development.

**Table 1. Participant Characteristics**

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Sibs-ASD (n = 64)</th>
<th>Sibs-TD (n = 42)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Chronological age, mo</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mean (SD)</td>
<td>16.3 (3.8)</td>
<td>16.2 (3.4)</td>
</tr>
<tr>
<td>Range</td>
<td>12-23</td>
<td>12-23</td>
</tr>
<tr>
<td>Sex</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>35 (55)</td>
<td>25 (60)</td>
</tr>
<tr>
<td>Female</td>
<td>29 (45)</td>
<td>17 (40)</td>
</tr>
<tr>
<td>Race</td>
<td></td>
<td></td>
</tr>
<tr>
<td>White</td>
<td>53 (83)</td>
<td>39 (93)</td>
</tr>
<tr>
<td>African American</td>
<td>4 (6)</td>
<td>0</td>
</tr>
<tr>
<td>Other</td>
<td>7 (11)</td>
<td>3 (7)</td>
</tr>
<tr>
<td>Maternal education</td>
<td></td>
<td></td>
</tr>
<tr>
<td>High school diploma</td>
<td>64 (100)</td>
<td>41 (98)</td>
</tr>
<tr>
<td>College degree</td>
<td>40 (62)</td>
<td>37 (88)</td>
</tr>
<tr>
<td>Hollingshead prestige score†</td>
<td>51.5 (11.1)</td>
<td>54.5 (10.0)</td>
</tr>
<tr>
<td>Range</td>
<td>29-66</td>
<td>26-66</td>
</tr>
</tbody>
</table>

Abbreviations: Sibs-ASD, younger siblings of children with an autism spectrum disorder; Sibs-TD, siblings of typically developing children. *Data are given as number (percentage) of each group unless otherwise indicated. †Scores range from 8 to 66, with higher scores reflecting higher socioeconomic levels.
Childhood Autism Rating Scale

The CARS\(^2\text{5}\) is a 15-item scale used to assess autism symptoms (eg, social relating, communication, and body use). Items are rated on a 4-point scale (including midpoints) according to degree of abnormality. Total scores range from 15 to 60, with scores 30 and higher suggesting the presence of autism. The CARS has strong test-retest reliability and correlations with clinical ratings.\(^2\text{6,27}\) This measure was completed collaboratively by the research team after observing the child’s behaviors during the entire session. The total CARS score was used as a continuous measure of autism symptoms, consistent with previous research.\(^2\text{6,27}\)

Screening Tool for Autism in Two-Year-Olds

The Screening Tool for Autism in Two-Year-Olds (STAT)\(^2\text{8,29}\) is an interactive screening tool developed to identify autism risk in children between the ages of 24 and 36 months. It consists of 12 activity-based items that are coded live and assess 4 social-communicative domains: play (2 items), directing attention (4 items), and motor imitation (4 items). Within each domain, items are scored as passed or failed according to specific behavioral criteria, and domain scores reflect the number of items passed. The total STAT score is calculated from the average number of failures across domains; this score ranges from 0 to 4 (in increments of 0.25). Higher total scores represent more impaired social-communicative performance, with scores of 2 or greater indicating autism risk. The STAT has strong screening properties for those aged 24 to 36 months, including sensitivity and specificity, interobserver agreement, test-retest reliability, and concurrent validity with the Autism Diagnostic Observation Schedule and clinical diagnosis.\(^2\text{9}\) Prior research\(^3\text{0}\) has used the STAT in children younger than 24 months, and preliminary data have revealed strong psychometric properties for children as young as 14 months, using a higher cutoff score (ie, 2.75) (W.L.S. and L. Henderson, PhD, unpublished data, 2006). In the present study, the total STAT score was used as a measure of overall social-communicative functioning, and the 4 individual domain scores were used to measure specific social-communicative skills.

MacArthur Communicative Development Inventories

The MacArthur Communicative Development Inventories (MCDI)\(^3\text{0}\) is a parental report measure of child language and communication development commonly used with young children with ASD.\(^3\text{1}\) Four scores from the Words and Gestures form were used: number of words understood, number of words used, number of phrases understood, and number of communicative gestures used. Vocabulary scores were derived by summing the number of words on a 396-item vocabulary checklist endorsed as understood (vocabulary comprehension) or understood and used (vocabulary production). The score for gestures used was derived from a 12-item scale with a 3-point response format (ie, not yet, sometimes, or often); the latter 2 responses were collapsed into a single category for analysis. The score for phrases understood was derived by summing the number of items endorsed from a list of 28 phrases.

Detection of Autism by Infant Sociability Interview

The Detection of Autism by Infant Sociability Interview (DAISI)\(^3\text{2}\) is a semistructured parental interview developed to measure social engagement behaviors present before the age of 2 years. Items assess early dyadic (eg, turn taking) and triadic (eg, referential eye contact) interactions. Retrospective use of the DAISI with parents of undiagnosed 2- to 4-year-old children found that 15 key items differentiated children who were subsequently diagnosed as having autism from those diagnosed as having nonautistic developmental delay.\(^3\text{1}\) In the present study, the 15-item version was used; scores range from 0 to 15, with higher scores reflecting more optimal social-communicative development.

RESULTS

PRELIMINARY ANALYSES

Preliminary analyses were conducted to examine the relationship between maternal education and chronological age and the other variables of interest. When appropriate, maternal education, chronological age, or both were entered as covariates in subsequent analyses. Mean differences and 95% CIs are reported for analyses of covariance and multivariate analyses of covariance. Results from \(\chi^2\) analyses are presented as odds ratios (ORs) (Sibs-ASD/Sibs-TD) with 95% CIs.

CHILD-BASED MEASURES

Children in the Sibs-ASD group obtained significantly lower mean cognitive scores than those in the Sibs-TD group on the MSEL early learning composite (MD, 6.95; 95% CI, 1.45-12.45) and the MSEL visual reception domain (MD, 5.91; 95% CI, 2.48-9.34) (Table 2). Autism symptoms on the CARS were greater for Sibs-ASD than Sibs-TD (MD, 2.54; 95% CI, 1.05-4.03). In addition, social-communicative performance was lower for Sibs-ASD on the STAT total (MD, 0.31; 95% CI, 0.01-0.61) and the directing attention domain (MD, 0.52; 95% CI, 0.07-0.97).

The pattern of scores within each group suggested that the lower performance of the Sibs-ASD characterized a substantial subgroup of this sample, rather than a few outstanding low performers. For example, 34 (53%) of Sibs-ASD (vs 12 [29%] of Sibs-TD) obtained total STAT scores in the at-risk range (ie, \(\geq 2.75\)) (\(\chi^2=5.98\); OR, 0.30 [95% CI, 0.11-0.81]).

PARENTAL REPORT MEASURES

Significant group differences were found for the DAISI (MD, 1.32; 95% CI, 0.27-2.37), with parents of Sibs-ASD reporting fewer social-communicative behaviors. On the MCDI, significant differences were found for vocabulary comprehension (MD, 33.30; 95% CI, 3.11-63.48), phrases understood (MD, 4.56; 95% CI, 1.85-7.27), and gestures understood (MD, 1.49; 95% CI, 0.51-2.47) (Table 3). Again, a substantial minority of Sibs-ASD were in the lower-performing subgroup. For example, 19 (30%) Sibs-ASD (vs 1 [2%] Sibs-TD) reportedly understood fewer than 10 phrases (\(\chi^2=12.61\); OR, 17.71 [95% CI, 2.27-138.27]), 10 (16%) (vs 0) used fewer than 5 gestures (\(\chi^2=7.00\); OR, undefined), 35 (55%) (vs 14 [33%]) understood fewer than 100 words (\(\chi^2=5.00\); OR, 2.50 [95% CI, 1.11-5.63]), and 9 (14%) (vs 0) obtained DAISI scores...
lower than those previously reported for children with developmental delay (χ² = 6.45; OR, undefined).32

Correlations between child-based and parental report variables measuring similar constructs revealed significant agreement. Parental report on the DAISI was correlated with children’s total STAT and CARS scores, and MSEL language scores were correlated with MCDI scores (Table 4).

DISTRIBUTIONS OF SCORES

Scatterplots were used to examine patterns of scores within and between groups. Figure 1 illustrates patterns of performance on the STAT and CARS, both of which measure autism symptoms. For most Sibs-TD (36 [86%] of 42 children), scores clustered in the lower left quadrant, indicating few or no signs of autism symptoms. Scores for the Sibs-ASD showed more variability; whereas most (41 [64%] of 64) children demonstrated a pattern similar to the Sibs-TD, a substantial minority of the Sibs-ASD obtained scores in the at-risk range (ie, ≥2.75) on the STAT (23 [36%] of 64 children), 3 of whom also had clinical scores (ie, ≥30) on the CARS. Six of the Sibs-TD also obtained scores in the at-risk range on the STAT; however, 5 of these children were younger than 14 months, which is below the age for which it is recommended. None of the Sibs-TD scored in the clinical range on the CARS.

Figure 2 provides a comparison of patterns across parental report (DAISI) and child-based (STAT) measures of social-communicative behaviors. Again, many of the Sibs-TD (36 [86%] of 42 children) demonstrated well-developed social-communicative skills (ie, higher DAISI scores and lower STAT scores). The Sibs-ASD demonstrated more variable performance, with higher proportions obtaining suboptimal scores on 1 (16 [25%] of 64 children) or both (8 [12%] of 64 children) measures.

The results of this study revealed that Sibs-ASD demonstrated significantly lower performance across measures of social-communicative development, cognitive functioning, and autism symptoms, relative to their low-risk peers. Weaker social-communicative performance for Sibs-ASD was found on the STAT, an interactive measure assessing play, imitation, and communication; the DAISI, a parent interview assessing early social, affective, and communication behaviors; and the MCDI, a parental questionnaire assessing verbal and nonverbal understanding and expression. The consistency of results obtained across different methods highlights the robustness of these findings.

Results of cognitive testing with the MSEL revealed group differences for the composite score and for the scale
assessing nonverbal problem solving (ie, visual reception). However, mean scores for both groups were well within the average range. The weaker performance on visual reception demonstrated by the Sibs-ASD was somewhat surprising, given that nonverbal ability is often described as a relative strength for young children with ASD.33 However, several items on this scale involve the presentation of verbal instructions and/or require a gestural response (eg, pointing). It is, thus, possible that these findings are related to the lower language understanding and gesture use demonstrated by Sibs-ASD. Significant positive correlations between the MSEL and MCDI support this interpretation.

Our failure to find expressive language differences between the sibling groups, either for parental report or cognitive measures, was also unexpected. Language impair-

**Table 4. Correlations Between Child-Based and Parental Report Measures**

<table>
<thead>
<tr>
<th>Child-Based Measure</th>
<th>DAISI Total</th>
<th>Vocabulary Production</th>
<th>Gestures Used</th>
<th>Vocabulary Comprehension</th>
<th>Phrases Understood</th>
</tr>
</thead>
<tbody>
<tr>
<td>STAT</td>
<td>-0.37*</td>
<td>-0.46*</td>
<td>-0.61*</td>
<td>-0.51*</td>
<td>-0.50*</td>
</tr>
<tr>
<td>Play</td>
<td>0.26*</td>
<td>0.30*</td>
<td>0.42*</td>
<td>0.40*</td>
<td>0.38*</td>
</tr>
<tr>
<td>Requesting</td>
<td>0.25†</td>
<td>0.28*</td>
<td>0.45*</td>
<td>0.37*</td>
<td>0.39*</td>
</tr>
<tr>
<td>Directing attention</td>
<td>0.17</td>
<td>0.33*</td>
<td>0.37*</td>
<td>0.26*</td>
<td>0.26*</td>
</tr>
<tr>
<td>Imitation</td>
<td>0.29*</td>
<td>0.29*</td>
<td>0.33*</td>
<td>0.27*</td>
<td>0.23†</td>
</tr>
<tr>
<td>MSEL</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>ELC</td>
<td>0.46*</td>
<td>0.25†</td>
<td>0.47*</td>
<td>0.30*</td>
<td>0.38*</td>
</tr>
<tr>
<td>Visual reception</td>
<td>0.28*</td>
<td>0.09</td>
<td>0.25†</td>
<td>0.15</td>
<td>0.23†</td>
</tr>
<tr>
<td>Fine motor</td>
<td>0.22†</td>
<td>-0.11</td>
<td>0.15</td>
<td>-0.11</td>
<td>-0.03</td>
</tr>
<tr>
<td>Receptive language</td>
<td>0.41*</td>
<td>0.33*</td>
<td>0.47†</td>
<td>0.46*</td>
<td>0.53*</td>
</tr>
<tr>
<td>Expressive language</td>
<td>0.39*</td>
<td>0.30*</td>
<td>0.40*</td>
<td>0.25*</td>
<td>0.27*</td>
</tr>
<tr>
<td>CARS total</td>
<td>-0.74*</td>
<td>-0.06</td>
<td>-0.46*</td>
<td>-0.21†</td>
<td>-0.30*</td>
</tr>
</tbody>
</table>

Abbreviations: CARS, Childhood Autism Rating Scale; DAISI, Detection of Autism by Infant Sociability Interview; ELC, early learning composite; MCDI, MacArthur Communicative Development Inventories; MSEL, Mullen Scales of Early Learning; STAT, Screening Tool for Autism in Two-Year-Olds.

*P<.01.
†P<.05.
ments, including early delays in spoken language, are a prominent feature of autism and an important behavioral marker for early identification.47-50 This finding may be explained by the young ages of the children in this study. Similar results with young ASD group siblings were obtained by Mitchell et al.,13 who concluded that gestural deficits may be apparent earlier than language deficits and may, therefore, represent more sensitive markers for early detection.

Group differences for directing attention (ie, initiating joint attention), which represents a core feature of autism,36-38 were found on the STAT. This result is consistent with findings from other recent studies,14,21,39,40 demonstrating differences in initiating joint attention for Sibs-ASD on the Early Social Communication Scale. Joint attention is a key developmental skill that has been associated with language acquisition in children with typical development and in those with autism.41-46 The implications of early developmental differences in joint attention for subsequent language development in Sibs-ASD are not yet known, because normative data are not available for many social-communicative behaviors, including joint attention. As a result, the point at which behavioral differences represent delayed or disordered development (or risk for such) is not known.

One important question regarding group comparisons between siblings of children with ASD and siblings of children with typical development is the extent to which group differences may be attributable to a few Sibs-ASD whose performance may be particularly impaired. Our results suggest that a substantial proportion of Sibs-ASD—rather than a few “outliers”—demonstrated weaker performance relative to the Sibs-TD. For example, more than half of the Sibs-ASD obtained 1 or more below-average MSEL subtest scores and more than one third obtained STAT scores in the “at-risk” range. Moreover, scatterplots illustrated the clustering of scores within the optimal range for the Sibs-TD, whereas the Sibs-ASD demonstrated considerable variability in performance. These results are consistent with those of Goldberg et al.,21 who found a wide distribution of Early Social Communication Scale scores in a young sample of Sibs-ASD.

Because the study of infant siblings is a fairly new area of research, follow-up diagnostic evaluations have extended only to the age of 24 months. Thus, we have little information about long-term developmental pathways or trajectories. It could be the case that mild behavioral differences in this group resolve on their own over time, without intervention and with no ill effects. On the other hand, it is possible that slight differences in social-communicative behavior may limit a young child’s exposure to crucial social input and result in a cascade of developmental sequelae.37 Conceptualizations of brain development suggest that atypical early experiences may have a significant effect on brain growth and later neuropsychological functioning.47-50 Therefore, understanding the early development of these high-risk siblings may suggest treatment (or prevention) strategies for attenuating early developmental differences and optimizing outcomes. Longitudinal follow-up of these high-risk siblings is under way, and will clarify the implications of early behavioral differences. Meanwhile, however, developmental surveillance is important to ensure that any needed assessment or intervention referrals are made as early as possible.

In sum, the study of infant siblings of children with ASD offers an important opportunity to learn about the earliest signs of autism and/or broader phenotypic features, and to understand normative and disrupted patterns of early social-communicative development. This research has the potential to increase our knowledge about the early development of autism and to develop tailored intervention and prevention strategies for promoting optimal outcomes in this group of at-risk children.

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REFERENCES


