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ORIGINAL PAPER

Early Social, Imitation, Play, and Language Abilities of Young Non-Autistic Siblings of Children with Autism

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Abstract Studies are needed to better understand the broad autism phenotype in young siblings of children with autism. Cognitive, adaptive, social, imitation, play, and language abilities were examined in 42 non-autistic siblings and 20 toddlers with no family history of autism, ages 18–27 months. Siblings, as a group, were below average in expressive language and composite IQ, had lower mean receptive language, adaptive behavior, and social communication skills, and used fewer words, distal gestures, and responsive social smiles than comparison children. Additionally, parents reported social impairments in siblings by 13 months of age. These results suggest that the development of young non-autistic siblings is affected at an early age and, thus, should be closely monitored, with appropriate interventions implemented as needed.

Keywords Autism · Siblings · Social · Imitation · Play · Language

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Introduction

Autism is a severe, neurodevelopmental disorder characterized by impairments in social and communication behaviors, and a restricted range of activities and interests. Once considered rare, autism is now believed to affect as many as 3 in 500 individuals (Baird et al., 2000; Bertrand et al., 2001; Chakrabarti & Fombonne, 2001; Fombonne, 2003). Historically, individuals with autism have had very poor prognoses; however, more recent research suggests that early intensive intervention can lead to improved outcomes for many individuals with autism spectrum disorders (Birnbrauer & Leach, 1993; Dawson & Osterling, 1997; McEachin, Smith, & Lovaas, 1993; Rogers, 1998; Sheinkopf & Siegel, 1998). Thus, there is a critical need for research that aims to improve methods of early detection of infants and toddlers at risk for autism. Furthermore, it is now recognized that a familial-genetic basis for autism exists, evidenced by an increased concordance for autism in monozygotic twins (Bailey, LeCouteur, Gottesman, & Bolton, 1995; Folstein & Rutter, 1977), and an increased risk of recurrence of autism in families who already have a child with autism (Jorde et al., 1990, 1991). There is also now evidence for a behavioral phenotype in relatives (parents, siblings) that is milder but qualitatively similar to the defining features of autism, referred to as the broad autism phenotype (Baron-Cohen & Hammer, 1997; Bolton et al., 1994; Bolton & Rutter, 1990; Starr et al., 2001).

A number of prospective studies focusing on infant siblings of children with autism are currently being conducted to better understand both the early development of autism and the broad autism phenotype in non-autistic siblings. The current study sought to add to this literature by examining a broad range of skill domains—social, imitation, play, and language—in 18- to 27-month-old non-autistic siblings of children with autism as compared to toddlers with no family history of autism. This information will help to alert parents to the kinds of difficulties that are likely to be manifest in siblings of children with autism. By recognizing these broad phenotype characteristics as early as possible, parents will be able to seek appropriate early intervention for siblings. Furthermore, such characteristics can offer clues to what facets of the autism syndrome tend to aggregate in families, which is useful for genetic studies.

A few published studies to date have reported on the developmental profiles of non-autistic siblings of children with autism under age two. In a recently published study of 21 siblings of children with autism and 21 siblings of typically developing children with no family history of autism, Yirmiya and colleagues (2006) found that, as early as 4 months, dyads of mothers and siblings of children with autism were less synchronous during mother–child interactions led by the infant, and the siblings were less upset by a still-face paradigm, as compared to dyads and sibs of typical children. Surprisingly, however, siblings of children with autism responded to their name being called more often than sibs of typical children at 4 months. At 14 months, sibs of children with autism were found to use requesting gestures less often, and had lower language scores, than sibs of typical children. In another recent study, Goldberg and colleagues (2005) found that 14–19 month old non-autistic siblings of children with autism obtained lower scores on the Early Social and Communication Scales (ESCS; Mundy, Delgado, Hogan, & Doehring, 2003; Seibert & Hogan, 1982) as compared to toddlers with typical development. Non-autistic siblings actually performed similarly to children with autism on measures of responding to social interaction, initiating joint attention, and requesting behavior. However, this study was limited by a very small sample size (8 children with autism, 8 siblings, and 9 typically developing children). Landa and Garrett-Mayer (2006) used the Mullen Scales of Early Learning (Mullen, 1997) to assess 60 siblings of children with autism and 27 age-matched low risk (i.e., no family history of autism) toddlers at 6, 14, and 24 months of age. The children were classified at 24 months of age into three groups: children with an ASD (22 siblings and 2 low risk), children with language delay (LD; 9 siblings and 2 low risk), and unaffected children (29 siblings and 23 low risk). Results showed that, at 14 months, the ASD group had lower scores in all Mullen domains than the unaffected group, and the LD group (largely composed of non-autistic siblings) had lower fine motor and receptive language skills than the unaffected group. By 24 months, the LD group performed similarly to the unaffected group in motor skills, but continued to show lower scores in visual problem solving and receptive and expressive language skills. At 14 months, the LD group also showed a pattern of higher visual versus receptive language skills, similar to the ASD group, although this difference was no longer apparent at 24 months of age for the LD group. In a study by Zwaigenbaum and colleagues (2005), 65 siblings of children with autism were compared to 23 low-risk (i.e., no family history of autism) infants from 6 to 24 months of age. Although differences were found primarily between siblings who later went on to meet criteria for ASD versus non-autistic siblings and comparison children, non-autistic siblings were found to have lower receptive language scores and used fewer phrases and gestures than comparison children at 12 months. In addition, latency to disengage attention became longer between 6 and 12 months of age for siblings, although this was particularly true for siblings who later met criteria for autism.

A number of studies focusing on language abilities in young non-autistic siblings have also been conducted. Ozonoff, Rogers, and Sigman (2005) recently reported on a large sample of toddlers (86 sibs of children with autism and 37 sibs of children with typical development). They found that at 18 months, siblings of children with autism obtained lower receptive and expressive language scores on the Mullen Scales of Early Learning (Mullen, 1997) compared to sibs of typically developing children. However, in this study, 36-month data had not yet been obtained and so it is not clear if the group means were lowered primarily by siblings who later were diagnosed with autism. In another study that followed infant siblings from 4 to 54 months of age, Yirmiya and colleagues (2005) reported on the cognitive and verbal abilities of 30 non-autistic siblings of children with autism as compared to 30 siblings of typically developing children. At 14 months, non-autistic siblings of children with autism had lower language scores on the Bayley Scales of Infant Development (Bayley, 1993) than sibs of typically developing children. However, by 24 months of age, there were no group differences on the Bayley or the Reynell Developmental Language Scales (Reynell & Gruber, 1990), although a significant number of siblings of children with autism scored 1 and 2 SDs below the mean in receptive language skills as compared to sibs of typically developing children. At 36 months, siblings of children with autism had lower receptive language scores on the Clinical Evaluation of Language Fundamentals-Preschool (CELF-P; Wiig, Secord, & Semel, 1992) compared to sibs of typical children, but by 54 months, there were no overall group differences (although a greater number of siblings of children with autism scored 1 SD below the mean as compared to siblings of typical children).

In sum, there have been only a few studies to date with adequate sample sizes that have focused on the early behaviors of non-autistic siblings. The current study represents a unique contribution to the literature by using multiple measures to examine a broad range of early abilities—cognitive, social communication, social-emotional functioning, imitation, functional and symbolic play, and language skills—in 18 to 27-month-old non-autistic siblings of children with autism as compared to toddlers with no family history of autism. Further, detailed data on children's development from birth to two years of age were obtained by parent interview. Finally, multiple measures were used to assess parent mental health, marital relationship, and stress to determine whether differences in child behavior could be accounted for by differences in parental well-being.

Method

Participants

Participants were 42 non-autistic siblings of children with autism (*M* CA = 20.31 mos., *SD* = 2.18, range 18–25 mos., 21 M, 21 F), who are participating in a larger, ongoing NIMH-funded early detection study of children at high-risk for developing autism (i.e., infant siblings of children with autism), and 20 typically developing children with no family history of autism (11 had older siblings, 9 did not; *M* CA = 22.4 mos., *SD* = 2.80, range 19–27 mos., 13 M, 7 F), who are participating as a comparison group in an ongoing NIMH-funded study of early characteristics of autism (Dawson, PI). A Pearson chi-square test confirmed that the gender ratio for the two groups was non-significant (χ^2 (1, N = 62) = 1.231, p = .267). The two groups differed, on average, by 2 months of age (F(1, 61) = 10.33, p < .01); therefore, CA was entered as a covariate in all analyses. Ethnicity for the sibling group: 28 European American, 9 Multiracial, 4 Asian/Pacific Islander, and 1 Native American; for the comparison group: 18 European American, 1 Multiracial, and 1 African American.

Participants were recruited from the Center on Human Development and Disability (CHDD), the University of Washington Autism Center, local public health agencies, pediatric offices, and Birth-to-Three Centers. Exclusionary criteria included the presence of a neurological disorder of known etiology (e.g., Down syndrome, fragile X syndrome), significant sensory or motor impairment, major physical/medical problems, psychoactive medication, or history of serious head injury and/or neurological disease. Children in the comparison group were excluded if they exhibited accelerated development (a score of >115) as assessed by the Mullen Scales of Early Learning (Mullen, 1997) or if they had a family history of autism. The justification for this is that there tends to be a selection bias in university-recruited samples and we did not want to create artificial differences by having a comparison group that was above average in IQ. Over the course of the study, 13 children were excluded from the comparison group based on above average composite IQ scores on the Mullen.

Measures

The following measures were collected over 3 days of testing. For all tests the child sat in the parent's lap and breaks were given as needed. The clinicians administering these measures were not blind to group membership.

Diagnostic Assessment

All children were administered a standardized diagnostic assessment, and siblings meeting criteria for Autistic Disorder or Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS) (n = 12 from a larger sample of 62 children) were excluded from the final sample. The diagnostic assessment consisted of: (1) the Autism Diagnostic Interview—Revised, Toddler version (ADI-R; Lord, Rutter, & Le Couteur, 1994), which was administered only to the sibling group, (2) the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DiLavore, & Risi, 1999; Lord et al., 2000), administered to all children, and (3) clinical judgment of an experienced clinician based on presence/absence of autism symptoms per the Diagnostic and Statistical Manual of Mental Disorders—Fourth Edition (DSM-IV; American Psychiatric Association, 1994), completed for all children. Diagnosis of autism was defined as meeting criteria for Autistic Disorder on the ADOS and ADI-R and meeting DSM-IV criteria for Autistic Disorder based on clinical judgment. In addition, if a child received a diagnosis of Autistic Disorder on the ADOS and based on DSM-IV clinical diagnosis, and came within 2 points of meeting criteria on the social or communication domains of the ADI-R, the child was also considered to have Autistic Disorder. Diagnosis of PDD-NOS was defined as meeting criteria for PDD-NOS on the ADOS, meeting criteria for Autistic Disorder on the ADI-R or missing criteria on the social or communication domains of the ADI-R by 2 or fewer points, and meeting DSM-IV criteria for PDD-NOS based on clinical judgment. Children in the comparison group were administered the ADOS to ensure that they did not meet criteria for Autistic Disorder or PDD-NOS on the ADOS or based on clinical judgment, and did not show elevated symptoms on these measures. Additionally, all older siblings with autism in the study were administered the same diagnostic assessment battery as younger siblings. Of 62 children in the larger sample, 8 were excluded because the older sibling did not meet criteria on all three diagnostic instruments as described above.

Cognitive, Language, Motor, and Adaptive Behavior

The Mullen Scales of Early Learning (Mullen, 1997) is a standardized measure for use with infants and preschool children from birth through 68 months. The Mullen assesses gross motor, visual reception, fine motor, receptive language, and expressive language abilities, and also yields a composite score. The Vineland Adaptive Behavior Scales (Sparrow, Balla, & Cicchetti, 1984), a well-standardized measure of adaptive functioning, was administered to all children.

Social-emotional Functioning

The Vineland Social-Emotional Early Childhood Scales (SEEC; Sparrow, Balla, & Cicchetti, 1998), which assesses social-emotional functioning in early childhood and yields domain scores—including Interpersonal Relationships, Play and Leisure Time—as well as a composite score, was administered to all children. The Vineland SEEC contains all items from the Vineland socialization domain as well as additional items, thereby yielding a more comprehensive measure of social-emotional functioning than the Vineland alone.

Social Communication

The Communication and Symbolic Behavior Scale-Developmental Profile (CSBS-DP; Wetherby & Prizant, 1998, 2002) was used to sample and rate a variety of communicative and symbolic behaviors. The child sat at a table next to an examiner and parent. Two posters hung on the walls of the room. One at a time, the child was presented with a variety of toys, including a wind-up toy, balloon, bubbles, a jar filled with cereal, a bag of toys, books, a Kermit doll, toy kitchen set, and blocks. Twice, the examiner gained the child's attention and pointed to one of the posters saying, ''Look.'' Behaviors were rated from videotape by a clinician trained and reliable on this measure (i.e., intraclass correlation coefficients of .90 or greater for each item). Dependent variables included gaze shifts, shared positive affect, gaze/point following (responding to joint attention), rate of communicating, behavior regulation (requesting), social interaction, initiating joint attention, conventional gestures (pushing objects away, showing objects, pointing to objects), distal (the child's hand did not touch the object) gestures, sounds, words, language comprehension, functional and symbolic object use, and three composite scores (social, speech, symbolic). The concurrent and predictive validity of the CSBS-DP has been well established (Wetherby, Allen, Cleary, Kublin, & Goldstein, 2002; Wetherby & Prizant, 1998, 2002).

Imitation

Immediate and deferred motor imitation abilities were assessed using a battery developed by Meltzoff (Meltzoff, 1988; Rast & Meltzoff, 1995) consisting of 10 immediate and 5 deferred imitation tasks. A range of tasks was used, including body movements (e.g., eye-blinking), novel acts on objects (e.g., touching elbow to a panel), and familiar acts on objects (e.g., banging wooden blocks). The tasks were administered with the child sitting across from an examiner at a small table. After gaining the child's attention, the experimenter demonstrated each target act three times in 20 s. No verbal description of the tasks, and no physical prompting of the child, was given. For deferred imitation items, the examiner demonstrated all 5 target acts and then a 10-min delay was imposed, after which the child was presented with the test objects one at a time in their original order. Responses were coded from videotape by a primary and reliability coder (20% of the sample). Intra- and inter-scorer agreement exceeded $r = .90$. Dependent variables included the total number of target acts performed (possible range of 0–15), number of immediate imitation acts (0–10), and number of deferred imitation acts (0–5).

Play

The Play Assessment Scale (PAS; Fewell, 1992) was used to assess functional and symbolic play skills and contains 31 items ordered developmentally, designed for use with children 2–30 months. The child was presented with a variety of play materials and the child's functional and symbolic use of the objects was scored as follows. Raw scores were calculated based on basal (three consecutive 4-point scores) and ceiling (three consecutive scores of 0) performances, with 5 points possible for each item: spontaneous play (4 points), after a general verbal prompt (e.g., ''What can you do with these?'') (3 points), after a specific verbal instruction (e.g., ''Feed the doll'') (2 points), after a specific verbal instruction with the examiner modeling the action (1 point), or the child did not engage in the target action (0 points).

Parent Report of Early Development

The Early Development Interview (EDI; Werner, Dawson, & Munson, 2001; Werner, Dawson, Munson, & Osterling, 2005) is a 96-item parent-report interview that incorporates mnemonic techniques to improve accuracy of parent recall and inquires about symptoms in four domains (social, communication, regulatory/sensory, and repetitive) from birth to two. Items assess eye contact, orienting to social cues, interest in reciprocal social games, communicative babbling, joint attention, and atypical sensory/motor behaviors, among others. The EDI also measures family changes (e.g., birth of a sibling, move to a new home), child medical events (e.g., high fevers, head injuries), and stressors during pregnancy (e.g., financial/employment problems, diagnosis of older child). Children with autism versus DD versus typical development have been shown to significantly differ on the EDI by 12–15 months (Werner et al., 2005).

Parental Mental Health, Marital Relationship, and Stress

Three measures of parental functioning were administered to both mothers and fathers. The Brief Symptom Inventory (BSI), a 53-item self-report scale, measures nine symptom dimensions (somatization, obsessive-compulsive behavior, interpersonal sensitivity, depression, anxiety, hostility, phobic anxiety, paranoid ideation, and psychoticism), and yields three global indices (Global Severity Index (GSI), Positive Symptom Distress Index (PSDI), and Positive Symptom Total (PST)) (Derogatis, 1975). The Dyadic Adjustment Scale (DAS), a 32-item self-report measure, assesses the quality of the current relationship (Spanier, 1976). The Life Experiences Survey (LES), a 57-item self-report measure, assesses significant events experienced during the preceding 12 months, and the extent to which each event had a positive or negative impact on one's life (Sarason, Johnson, & Siegal, 1978).

Results

All children and parents with complete data were included in analyses. Mean differences were assessed using chronological age as a covariate, except when standard scores were examined (Vineland measures, Mullen Scales of Early Learning), as these already correct for chronological age. Tables 1–5 show the means, standard deviations (or standard errors), effect sizes (Cohen's D), and statistical results for each of the variables examined.

Cognitive and Adaptive Behavior

Multivariate analysis of variance was initially used to test for group differences in developmental level (cognitive functioning or IQ) as assessed by the Mullen Scales of Early Learning. However, the MANOVA omnibus test was non-significant, possibly due to small sample size and inadequate power on this measure; therefore, separate univariate ANOVAs were used to examine group differences. Note that the Type I error rate was kept at .05 for each ANOVA, a liberal standard given the number of tests conducted. Results indicated that the two groups of children obtained comparable overall mean scores on composite IQ, visual reception, gross motor, and fine motor (see Table 1). Mean scores on each of these scales fell within the average range for both groups of children. However, 12 siblings (29% of the sibling group) had below average (i.e., below 85) composite IQ scores while none of the children in the comparison group scored below 85 in overall IQ. The Fisher exact test indicated that this difference was significant ($p = .005$). Receptive and expressive language scores are discussed below.

On the Vineland Adaptive Behavior Scales, results of a MANOVA indicated a significant group difference (Wilks' Lambda = .819, $F(5, 56) = 2.48$, $p = .042$), with significant mean differences on all domains except communication. On average, siblings scored below that of comparison children in daily living skills, socialization, motor skills, and on the overall adaptive behavior composite (see Table 1).

Language

Results of ANOVAs on the Mullen receptive and expressive language scores revealed a significant group

Table 1 Cognitive, motor, language, and adaptive functioning of siblings and comparison children

	Siblings		Comparison		ANOVA/MANOVA			
	M	*SD*	*M*	*SD*	*F*	df	*p*	*D*
Mullen[a]								
Composite IQ	99.05	16.94	100.80	6.34	0.20	1, 60	.657	0.14
Visual reception	51.50	8.50	48.90	7.49	1.37	1, 60	.247	0.32
Gross motor	45.46	6.27	45.80	8.50	0.03	1, 57[b]	.863	0.05
Fine motor	49.26	8.11	49.55	6.65	0.02	1, 60	.891	0.04
Receptive language	*46.55*	*15.60*	*54.50*	*6.40*	*4.78*	*1, 60*	*.033*	*0.67*
Expressive language	49.07	12.24	48.45	6.65	0.05	1, 60	.833	0.06
Vineland[c]								
Communication	95.24	14.53	100.90	7.32	2.70	1, 60	.106	0.50
Daily living skills	*85.83*	*7.27*	*92.90*	*8.32*	*11.67*	*1, 60*	*.001*	*0.91*
Socialization	*91.48*	*9.50*	*96.15*	*7.37*	*3.76*	*1, 60*	*.057*	*0.55*
Motor	*94.02*	*7.50*	*99.20*	*9.91*	*5.22*	*1, 60*	*.026*	*0.59*
Composite	*88.48*	*9.42*	*95.95*	*8.11*	*9.29*	*1, 60*	*.003*	*0.85*

Italics indicate significant differences

[a] On the Mullen, composite IQ scores between 85 and 115 are considered to be in the average range; for the subscales, *T*-scores between 40 and 60 are in the average range

[b] Fewer children completed the gross motor subtest of the Mullen

[c] On the Vineland, scores between 85 and 115 are in the average range

difference in receptive language, with siblings obtaining, on average, lower scores than children in the comparison group (see Table 1). The groups did not differ in mean expressive language ability. However, while the overall mean scores fell within the average range for both groups on these measures, 14 children (33%) in the sibling group had below average receptive language scores (a significant difference using the Fisher exact test, $p = .002$) and 8 (19%) had below average expressive language scores (Fisher exact test, $p = .035$). It should be noted that there was a significant correlation between receptive language and chronological age only for the sibling group ($r = .37$). CA was not related to any of the other scales of the Mullen for either group.

Social Communication

A MANCOVA revealed significant group differences on the CSBS-DP (Wilks' Lambda = .448, $F(22, 38) = 2.13$, $p = .02$), with siblings demonstrating fewer behaviors than comparison toddlers in rate of communicating and use of distal gestures (see Table 2). Siblings also obtained lower cluster scores in overall social use of gestures and words, and lower Social Composite scores, Symbolic Composite scores, which reflect frequency and variety of symbolic play as well as language comprehension, and Total Composite scores on this measure. The two groups did not differ in the frequency in which they engaged in the following behaviors: gaze shifts, shared positive affect, gaze/point following, behavior regulation, social interaction, joint attention, and use of conventional gestures. They also did not differ in overall Speech Composite scores.

On the ADOS, a MANCOVA revealed group differences (Wilks' Lambda = .513, $F(20, 40) = 1.90$, $p = .042$), with siblings responding to social smiles, and pointing to request and/or show objects to others, less often than children in the comparison group. There were no group differences in shared enjoyment, showing, initiating joint attention, and response to joint attention behaviors on this measure (see Table 2).

Social-emotional Functioning

On the Vineland SEEC, results of a MANOVA indicated a significant group difference (Wilks' Lambda = .784, $F(3, 58) = 5.31$, $p = .003$), with siblings obtaining significantly lower scores in the Interpersonal Relationships domain, and in the overall Social-Emotional Composite, as compared to comparison toddlers. The two groups did not differ, however, in the Play and Leisure Time domain (see Table 2).

Imitation

There were no significant group differences in immediate and deferred imitation, as measured by Meltzoff's imitation battery (Wilks' Lambda = .984, $F(2, 57) = .468$, $p = .629$; see Table 3).

Table 2 Social communication and social-emotional functioning of siblings and comparison children

	Siblings		Comparison		MANCOVA[a]			
	M[b]	*SE*	*M*	*SE*	*F*	df	*p*	*D*
CSBS-DP								
Individual items								
Gaze shifts	4.99	0.18	5.32	0.27	0.96	1, 59	.331	0.33
Shared positive affect	2.52	0.24	2.05	0.35	1.16	1, 59	.287	0.03
Gaze/point following	1.92	0.06	1.88	0.08	0.16	1, 59	.690	0
Rate of communicating	*15.74*	*0.36*	*17.34*	*0.53*	*5.85*	*1, 59*	*.019*	*0.88*
Behavior regulation	5.34	0.12	5.58	0.18	1.13	1, 59	.292	0.48
Social interaction	1.49	0.24	1.66	0.36	0.15	1, 59	.702	0.25
Joint attention	3.55	0.28	3.84	0.42	0.30	1, 59	.587	0.35
Conventional gestures	4.36	0.22	4.89	0.33	1.65	1, 59	.203	0.42
Distal gestures	*1.90*	*0.23*	*3.01*	*0.35*	*6.68*	*1, 59*	*.012*	*0.87*
Cluster scores								
Speech Words	*10.80*	*0.42*	*12.47*	*0.62*	*4.70*	*1, 59*	*.034*	*0.92*
Social:Gestures	*7.45*	*0.36*	*9.00*	*0.53*	*5.47*	*1, 59*	*.023*	*0.70*
Composite scores								
Social composite	*7.43*	*0.36*	*8.75*	*0.53*	*3.96*	*1, 59*	*.051*	*0.65*
Speech composite	10.52	0.41	11.87	0.62	3.10	1, 59	.083	0.83
Symbolic composite	*8.52*	*0.32*	*9.80*	*0.48*	*4.63*	*1, 59*	*.035*	*0.94*
Total composite	*92.67*	*1.97*	*101.34*	*2.93*	*5.64*	*1, 59*	*.021*	*1.02*
ADOS[c]								
Responsive social smile	*1.09*	*0.13*	*0.35*	*0.19*	*9.81*	*1, 59*	*.003*	*1.10*
Shared enjoyment	0.26	0.09	0.46	0.14	1.36	1, 59	.249	0.07
Showing	0.66	0.11	0.46	0.16	1.05	1, 59	.310	0.21
Initiating joint attention	0.69	0.13	0.64	0.19	0.05	1, 59	.831	0.05
Response to joint attention	0.29	0.12	0.14	0.17	0.54	1, 59	.464	0.44
Pointing	*1.18*	*0.13*	*0.63*	*0.19*	*5.48*	*1, 59*	*.023*	*0.03*
Vineland SEEC								
Interpers relationships	*85.67*	*9.82*	*93.10*	*11.08*	*7.14*	*1, 60*	*.010*	*0.71*
Play/Leisure time	93.64	8.10	96.60	8.38	1.77	1, 60	.189	0.36
Soc-Emot composite	*88.05*	*8.94*	*95.40*	*9.21*	*8.99*	*1, 60*	*.004*	*0.81*

Italics indicate significant differences

[a] CA was entered as a covariate, except for the Vineland SEEC analyses, which used standard scores

[b] Estimated marginal means and standard errors are reported for the CSBS-DP and ADOS; raw means and standard deviations are reported for the Vineland SEEC

[c] On the ADOS, lower scores reflect less impairment; scores range from 0 to 2

Play

There were no significant group differences in either functional or symbolic play skills as measured by the Play Assessment Scale (Wilks' Lambda = .947, $F(2, 56) = 1.58$, $p = .216$; see Table 3).

Parent Report of Early Development

Symptoms from the Early Development Interview from each 3-month time period (0–3 mos., 4–6 mos., 7–9 mos., 10–12 mos., 13–15 mos., 16–18 mos., and 19–24 mos.) were analyzed. Sensory items (i.e., sensitivity to noise and touch) were derived from the regulatory domain. Additionally, information on family changes, child medical events, and stressors during pregnancy were also analyzed. Separate univariate ANOVAs were used to test for group differences in these areas as well as the following behavior domains: social, communication, regulatory, sensory, and repetitive. Although several comparisons were made in these analyses, the alpha level was kept at .05 for each analysis as we were interested in each symptom domain at each time point (Perneger, 1998). A summary of results is presented in Table 4. Note that a number of items are included in each domain score, and the possible range of scores for each individual item is 0–2, with higher scores indicating greater impairment. Siblings were reported to show greater overall social difficulties, first apparent at 13–15 months of age, than typically developing children with no family history of autism. Siblings were also reported to show fewer regulatory difficulties (analysis of individual items revealed that these differences were due to differences in fussiness and sleep in particular) at 0–3 months of age as compared to children in the comparison group. Finally, children in the comparison group were reported to show a

Table 3 Imitation and play abilities of siblings and comparison children

	Siblings		Comparison		MANCOVA[a]			
	M[b]	*SE*	*M*	*SE*	*F*	df	*p*	*D*
Imitation								
Total[c]	9.25	0.59	8.20	0.89	0.88	1, 58	.352	0.15
Immediate	5.90	0.48	5.01	0.71	0.95	1, 58	.333	0.16
Deferred	3.35	0.18	3.18	0.27	0.24	1, 58	.625	0.07
Play								
Func. play	87.72	2.17	80.92	3.42	2.65	1, 57	.109	0.09
Symb. play	7.38	0.60	6.67	0.95	0.38	1, 57	.542	0.30

[a] CA was entered as a covariate

[b] Estimated marginal means and standard errors are reported

[c] A separate ANOVA was used for the Total variable, as this variable is the sum of the immediate and deferred scores

greater number of sensory behaviors (from 10–24 months) and repetitive behaviors (at 22–24 months) than siblings. There were no differences reported in family changes and child medical events during the first 2 years of life, nor in stressful events occurring during pregnancy, suggesting that these variables were unlikely to account for the differences found in the children's early behaviors.

Parent Mental Health, Marital Relationship, and Stress

Means and standard deviations for each variable and measure for mothers and fathers in the two groups are reported in Table 5. There were no significant group differences for mothers and fathers on any of the symptom dimensions or global indices of the Brief Symptom Inventory (for mothers: Wilks' Lambda = .939, $F(10, 35) = .23$, $p = .992$; for fathers: Wilks' Lambda = .702, $F(10, 25) = 1.06$, $p = .424$). On the Dyadic Adjustment Scale, the omnibus test indicated group differences for mothers (Wilks' Lambda = .727, $F(4, 38) = 3.58$, $p = .014$), but there were no group differences on any of the individual variables. Results for fathers indicated no group differences (Wilks' Lambda = .928, $F(4, 31) = .60$, $p = .667$). On the Life Experiences Survey, there were no group differences for mothers (Wilks' Lambda = .963, $F(4, 50) = .48$, $p = .749$) or for fathers (Wilks' Lambda = .955, $F(4, 43) = .51$, $p = .727$). These results suggest that levels of parental mental health, marital functioning, and stress as measured by these three questionnaires were essentially the same for both parents of comparison toddlers and parents of children with autism in this sample.

Discussion

A range of early social communication, imitation, play, and language abilities in 18- to 27-month-old non-autistic siblings of children with autism as compared to children with no family history of autism was examined to identify the kinds of impairments that may be evident early on in young siblings. Using multiple measures, both direct observation and parent report, we found that non-autistic siblings demonstrated a variable profile, with decrements in some aspects of social communication and language, but with intact skills in other areas. Specifically, we found that, on average, siblings had lower receptive language skills, lower adaptive behavior skills, and lower overall rates of social communication and social-emotional functioning than comparison children. In addition, a significant number of siblings demonstrated below average expressive language and overall cognitive abilities. Finally, siblings demonstrated fewer symbolic behaviors overall (a combination of language comprehension and symbolic object use during play), as well as less frequent use of words, distal gestures including pointing, and responsive social smiles during social interactions than comparison children. Since multiple comparisons were made without correction for family-wise error, it is possible that some of these findings achieved statistical significance by chance. However, in light of the fact that we actually excluded children who had above average IQ scores in the comparison sample, the finding of group differences in a number of areas can be considered a conservative result.

Further findings based on retrospective parent report indicated that siblings showed greater overall social difficulties, evident by 13 months of age, fewer early regulatory difficulties (i.e., less fussiness and fewer sleep problems at 0–3 months), and fewer sensory and repetitive behaviors during the second year of life than toddlers in the comparison group. This may reflect higher levels of passivity in siblings of children with autism. In autism, repetitive behaviors are often not yet present at this young of an age, and such behaviors have not been shown to reliably distinguish children with autism at such an early age. Perhaps this is true of non-ASD siblings as well. In certain other domains, such as imitation and play, and in other aspects of social communication, such as initiating and responding to joint attention, siblings performed similarly to typical children. In fact, the young siblings in this study were a heterogeneous group, and their performance on each of the measures ranged from below average to, in some cases, above average.

Table 4 Early symptoms in siblings and comparison children[a]

	Siblings		Comparison		ANOVA			
	M	*SD*	*M*	*SD*	*F*	df	*p*	*D*
0–3 months								
Regulatory (0–10)	*1.50*	*1.27*	*3.11*	*2.37*	*11.03*	*1, 54*	*.002*	*0.85*
Sensory (0–2)	0.03	0.16	0.11	0.47	1.00	1, 54	.322	0.23
4–6 months								
Regulatory (0–10)	1.16	1.13	1.44	1.79	0.53	1, 54	.468	0.19
Sensory (0–2)	0.00	0.00	0.11	0.47	2.16	1, 54	.148	0.33
Social (0–4)	0.08	0.36	0.17	0.51	0.55	1, 54	.462	0.20
7–9 months								
Regulatory (0–10)	1.08	1.08	1.50	1.20	1.74	1, 54	.193	0.37
Sensory (0–2)	0.05	0.23	0.22	0.65	2.11	1, 54	.152	0.35
Social (0–8)	0.24	0.59	0.17	0.51	0.19	1, 54	.667	0.13
Communication (0–2)	0.08	0.27	0.06	0.24	0.10	1, 54	.756	0.08
10–12 months								
Regulatory (0–2)	0.29	0.65	0.50	0.79	1.11	1, 54	.297	0.29
Sensory (0–4)	*0.13*	*0.41*	*0.50*	*0.79*	*5.32*	*1, 54*	*.025*	*0.59*
Social (0–8)	0.37	0.97	0.06	0.24	1.80	1, 54	.185	0.44
Communication (0–4)	0.32	0.96	0.22	0.55	0.15	1, 54	.703	0.13
Repetitive (0–2)	0.05	0.23	0.06	0.24	0.00	1, 54	.965	0.04
13–15 months								
Regulatory (0–2)	0.29	0.65	0.44	0.78	0.60	1, 54	.441	0.21
Sensory (0–2)	*0.08*	*0.27*	*0.44*	*0.78*	*6.67*	*1, 54*	*.013*	*0.62*
Social (0–14)	*1.68*	*2.28*	*0.50*	*0.86*	*4.52*	*1, 54*	*.038*	*0.69*
Communication (0–2)	0.45	0.80	0.28	0.67	0.61	1, 54	.438	0.23
Repetitive (0–2)	0.03	0.16	0.11	0.47	1.00	1, 54	.322	0.23
16–18 months								
Sensory (0–2)	*0.08*	*0.27*	*0.44*	*0.78*	*6.67*	*1, 54*	*.013*	*0.62*
Social (0–10)	*1.13*	*1.96*	*0.11*	*0.47*	*4.70*	*1, 54*	*.035*	*0.72*
Communication (0–6)	0.87	1.42	0.50	0.79	1.06	1, 54	.309	0.32
Repetitive (0–2)	0.08	0.27	0.11	0.47	0.10	1, 54	.748	0.08
19–21 months								
Sensory (0–2)	*0.08*	*0.27*	*0.50*	*0.79*	*8.82*	*1, 54*	*.004*	*0.71*
Social (0–10)	0.61	1.48	0.00	0.00	2.98	1, 54	.090	0.58
Communication (0–6)	0.58	1.03	0.50	0.86	0.08	1, 54	.779	0.08
Repetitive (0–2)	0.03	0.16	0.11	0.47	1.00	1, 54	.322	0.23
22–24 months								
Sensory (0–2)	*0.08*	*0.27*	*0.50*	*0.79*	*8.82*	*1, 54*	*.004*	*0.71*
Social (0–10)	0.32	0.74	0.00	0.00	3.25	1, 54	.077	0.61
Communication (0–8)	0.66	1.34	0.44	0.78	0.39	1, 54	.535	0.20
Repetitive (0–6)	*0.26*	*0.72*	*0.94*	*1.21*	*6.91*	*1, 54*	*.011*	*0.68*
Total scores								
Regulatory (0–34)	*4.32*	*3.60*	*7.00*	*4.59*	*5.68*	*1, 54*	*.021*	*0.65*
Sensory (0–18)	*0.53*	*1.57*	*2.83*	*4.61*	*7.77*	*1, 54*	*.007*	*0.67*
Social (0–64)	*4.42*	*6.60*	*1.00*	*1.68*	*4.66*	*1, 54*	*.035*	*0.71*
Communication (0–28)	2.95	4.32	2.00	2.57	0.74	1, 54	.395	0.27
Repetitive (0–14)	0.45	1.03	1.33	2.59	3.38	1, 54	.072	0.45
Family changes (0–18)	2.61	2.35	3.39	2.95	1.15	1, 54	.289	0.29
Child medical events (0–12)	1.87	1.86	2.00	1.82	0.06	1, 54	.804	0.07
Pregnancy stress events (0–16)	1.97	1.81	1.17	1.79	2.45	1, 54	.124	0.44

Italics indicate significant differences

[a] Higher scores on this measure indicate greater impairment

In terms of family environmental factors, parents of children with autism and parents of comparison children in this sample reported similar levels of mental health symptoms, marital adjustment and satisfaction, and stressful life events, suggesting that these parental factors were unlikely to account for the group differences found between comparison children and siblings.

The strengths of this study include the sample size (as compared to other published studies of young non-autistic siblings), a broader range of measures than has been used previously, and the use of multiple measures for many of the skill domains examined. Although several of our findings were consistent with those of previous studies, there were some skill domains, such

Table 5 Results of parent measures[a]

	Siblings		Comparison	
	Mother	Father	Mother	Father
BSI				
Anxiety	0.59 (0.56)	0.30 (0.28)	0.53 (0.48)	0.50 (0.68)
Depression	0.56 (0.77)	0.48 (0.62)	0.60 (0.69)	0.50 (0.98)
Hostility	0.71 (0.70)	0.50 (0.48)	0.64 (0.56)	0.69 (1.25)
Interpersonal sensitivity	0.66 (0.74)	0.28 (0.31)	0.63 (0.72)	0.39 (0.40)
OCD behavior	1.08 (0.77)	0.68 (0.68)	1.04 (0.71)	0.96 (0.78)
Paranoid ideation	0.47 (0.55)	0.44 (0.54)	0.47 (0.72)	0.60 (0.94)
Phobic anxiety	0.12 (0.30)	0.07 (0.14)	0.13 (0.22)	0.07 (0.14)
Psychoticism	0.32 (0.47)	0.24 (0.35)	0.28 (0.43)	0.51 (0.96)
Somatization	0.47 (0.55)	0.20 (0.32)	0.34 (0.37)	0.27 (0.44)
Positive symptom total	29.40 (26.10)	18.50 (16.27)	27.21 (22.20)	26.93 (35.93)
Global severity index	0.57 (0.50)	0.36 (0.31)	0.52 (0.43)	0.52 (0.69)
DAS				
Affectional expression	3.53 (1.50)	4.00 (1.62)	4.08 (1.26)	3.44 (1.42)
Dyadic cohesion	14.47 (3.07)	15.22 (2.93)	15.85 (3.16)	16.11 (2.52)
Dyadic consensus	14.50 (5.73)	15.52 (6.32)	14.08 (5.65)	16.00 (2.83)
Dyadic satisfaction	33.83 (4.14)	33.41 (4.08)	31.92 (6.25)	34.56 (3.50)
Total	66.33 (4.27)	68.15 (5.30)	65.92 (7.08)	70.11 (4.96)
LES				
Positive change	6.26 (6.03)	5.80 (5.67)	7.24 (6.84)	6.15 (7.95)
Negative change	6.87 (7.75)	5.94 (4.96)	5.88 (7.00)	5.15 (5.06)
Total change	–0.61 (7.83)	–0.14 (7.26)	1.35 (6.08)	1.00 (5.13)
# of positive events	3.13 (2.60)	3.11 (2.69)	3.71 (2.82)	2.92 (3.35)
# of negative events	3.55 (3.24)	3.86 (2.94)	3.41 (3.12)	3.31 (2.98)

[a] Numbers are means with standard deviations in parentheses

as joint attention, where we did not obtain similar results (e.g., Goldberg et al., 2005). There are several possible explanations for this. First, it is possible that some of our measures were less sensitive in capturing subtle differences. For instance, the CSBS-DP yields a more restricted range of scores for eye gaze, joint attention, behavior regulation, and social interaction than the Early Social Communication Scales (ESCS; Mundy, Delgado, Hogan, & Doehring, 2003; Seibert & Hogan, 1982), a comparable instrument that yields a greater range of frequency scores. Second, it is possible that, over time, many non-autistic siblings develop more frank difficulties that are not as evident at very young ages. For example, some of the impairments that have been observed in older siblings, including executive function deficits and verbal fluency impairments (Hughes, Plumet, & Leboyer, 1999), are not easily detected until children reach later preschool and school ages. Asperger syndrome is rarely identified in preschool, and symptoms of autism (e.g., poor joint attention, imitation, play) may not be apparent at that age or may not be part of the Asperger phenotype (see McConachie, Le Couteur, & Honey, 2005). Third, it is possible that previous studies have overestimated the number of non-autistic siblings that demonstrate broad phenotype characteristics. Many studies have relied on a family history interview method (e.g., Bolton et al., 1994) and have focused on older children. These studies require parents to report retrospectively on broad constructs, such as ‘‘impaired friendships.’’ Finally, the fact that prospective observations of younger siblings thus far have yielded mixed results likely reflects the variable presence of impairments in the sibling population at different ages as well as the types of measurements used across studies. For example, results may vary depending on whether data are collected by direct testing by experimenters in a laboratory setting versus parent report of behavior at home. Additionally, characteristics of the broad autism phenotype may vary across development, with some delays being transient while others are more lasting.

What might account for the differences found in siblings' early social, language, and adaptive functioning? Although this study did not address the question of causation, both genetic and environmental factors should be considered. Siblings are at heightened genetic risk for impairments due to the fact that they share genes in common with their sibling with autism. Added to this genetic vulnerability are environmental factors, including modeling of atypical behaviors by the sibling with autism, reduced opportunities for engaging in typical sibling social interactions, and possibly fewer interactions with parents whose child with autism may require high levels of attention. Sibling and parent–child interactions may not only be less frequent but also qualitatively different. It will be informative, in future studies, to examine early parent–child, and child–child, interactions as these play an important role in the development of early social and language abilities. Although these family factors may not cause the impairments found in siblings, it is possible that these factors may contribute additional risk (or protection) during a particular time in development (Bauminger & Yirmiya, 2001). For some siblings, the genetic involvement may be strong enough that under even the most optimal environment, these children develop impairments, while for other siblings, the genetic predisposition may be so weak that only under certain environmental conditions will these children exhibit the broader autism phenotype (Bauminger & Yirmiya, 2001, p. 75). This explanation is based on the diathesis-stress model and integrates both genetic and environmental factors in explaining development (Brown & Harros, 1989; Rende & Plomin, 1992, Walker, Downey, & Bergman, 1989, among others). According to this model, a genetic predisposition may be essential but not sufficient for the expression of a phenotype.

In summary, certain aspects of language, cognitive ability, adaptive behavior, and social functioning do appear to be affected at an early age in non-autistic siblings of children with autism and should be closely monitored by parents and professionals. In some cases, interventions, such as speech and language therapy and behavioral therapy approaches focusing on social skills, may be warranted. However, we are just now starting to learn about the early development of siblings of children with autism. Future research would benefit from the inclusion of comparison groups of developmentally delayed children, as well as siblings of children with other developmental disorders, to determine whether the early characteristics evidenced by siblings of children with autism are specific to families with a history of autism or are more generally related to families with children with special needs. In addition, young siblings should be followed over time to determine how these early characteristics change with development. Finally, additional environmental factors, including sibling interactions and parent–child interactions, should be considered.

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