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The development of siblings of children with autism at 4 and 14 months: social engagement, communication, and cognition

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Aims: To compare siblings of children with autism (SIBS-A) and siblings of children with typical development (SIBS-TD) at 4 and 14 months of age. **Methods:** At 4 months, mother–infant interactional synchrony during free play, infant gaze and affect during the still-face paradigm, and infant responsiveness to a name-calling paradigm were examined ($n = 21$ in each group). At 14 months, verbal and nonverbal communication skills were examined as well as cognition (30 SIBS-A and 31 SIBS-TD). **Results:** Most SIBS-A were functioning as well as the SIBS-TD at 4 and 14 months of age. However, some differences in early social engagement and later communicative and cognitive skills emerged. Synchrony was weaker in the SIBS-A dyads, but only for infant-led interactions. Infant SIBS-A revealed more neutral affect during the still-face procedure and were less upset by it than was true for the SIBS-TD. A surprising result was that significantly more SIBS-A responded to their name being called by their mothers compared to SIBS-TD. At 14 months, SIBS-A made fewer nonverbal requesting gestures and achieved lower language scores on the Bayley Scale. Six SIBS-A revealed a language delay of 5 months and were responsible for some of the significant differences between SIBS-A and SIBS-TD. Furthermore, infant SIBS-A who showed more neutral affect to the still face and were less able to respond to their name being called by their mothers initiated fewer nonverbal joint attention and requesting behaviors at 14 months, respectively. **Discussion:** Focused on the genetic liability for the broad phenotype of autism as well as the possible influence of having a sibling with autism. **Keywords:** Autism, siblings, still face, synchrony, mother–child interaction, language, nonverbal communication, broad phenotype, joint attention.

Siblings of children with autism (SIBS-A) are at greater risk for developing what is termed today the 'broad phenotype of autism', i.e., milder difficulties in one or more of the three areas that are impaired in autism: social responsiveness, communication, and limited interests/stereotyped behavior. An estimated 25% of siblings of people with autism show the broad phenotype of social and cognitive abnormalities, often in a subtle form. Family studies of autism also suggest an overlap between classic autism and other autism spectrum disorders including the broad phenotype (Pickles et al., 2000).

Researchers vary in how they operationalize the broad phenotype. Some employ standard diagnostic instruments such as the Autism Diagnostic Interview – Revised (ADI-R; Lord, Rutter, & Le Couteur, 1994) or other standardized tests (e.g., intelligence tests, reading and spelling tests) (Bolton et al., 1994), whereas others employ experimental tasks (e.g., Tower of Hanoi, theory of mind) (Baron-Cohen & Hammer, 1997; Happé, Briskman, & Frith, 2001). As a result, the broad phenotype has no single operational definition. Furthermore, some researchers report weaknesses, some report strengths, and some report non-significant findings (for review see Lainhart, 1999; Piven, 1999; Yirmiya, Shaked, & Erel, 2001). Other factors that may contribute to the inconsistent picture include the heterogeneity of siblings' ages in the various samples and the use of different comparison groups. Strikingly, most sibling studies are conducted in late childhood or even adolescence or adulthood, with almost none in infancy. Therefore studying young siblings of children with autism may be important because of the camouflaging effects of later compensation that may be operative in older siblings.

Based on the premise that social and communication difficulties may be among the best predictors for autism and thus for investigating the broad phenotype, and given the recent evidence that social behavior has a genetic component (Brothers, 1996; Constantino & Todd, 2000), the social responsiveness, communication, and cognition of SIBS-A at ages 4 and 14 months were investigated. No previous studies of siblings of various ages have included infant siblings or involved a longitudinal design (for a review, see Bauminger & Yirmiya, 2001; Yirmiya et al., 2001). The present study aimed to determine whether the development of these siblings at ages 4 and 14 months was delayed or deviant in areas of development which are known to be impaired in autism, compared to the development of SIBS-TD.

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However, we do not know how children with autism would score on the measures employed in the current study at age 4 or 14 months, because autism is typically not diagnosed before the second or third year of life.

At 4 months, SIBS-A and SIBS-TD were examined employing two well-established measures of social engagement. Social engagement itself has not been previously studied in young SIBS-A, yet researchers have reported that older SIBS-A reveal more behavior problems and less prosocial behavior compared to siblings of children with other diagnoses and/or to SIBS-TD (Hastings, 2003; Verte, Roeyers, & Buysee, 2003). Evidence of impaired social engagement in young infants who are later diagnosed with autism has been found from analysis of home movies provided by parents (Adrien et al., 1993; Baraneck, 1999; Osterling & Dawson, 1994; Werner, Dawson, Osterling, & Dinno, 2000). In these home-video studies, the social abnormalities first appeared at 8–18 months, which in one sense constitutes early signs but in another sense is relatively late in infancy. In addition, social engagement was evaluated using a variety of research methods so that questions remain about the measurements and ontogenesis of the social and other difficulties during the first year of life.

The social engagement measures included: (a) synchrony in mother–child face-to-face free interaction and (b) the still-face paradigm. A third measure, involving name-calling, was designed especially for the current study. Synchrony was assessed because of its possible association with the well-documented impairments in joint attention, social responsiveness, and theory of mind that characterize children with autism (Leekam, Baron-Cohen, Perret, Milders, & Brown, 1997; Sigman & Ruskin, 1999; Yirmiya, Erel, Shaked, & Solomonica-Levi, 1998). Synchrony has been previously examined in studies of normal and high-risk samples (Cohn & Tronick, 1988; Endriga & Speltz, 1997; Feldman, 2003; Field, Healy, Goldstein, & Guthertz, 1990; Lester, Hoffman, & Brazelton, 1985). Researchers found that mid-range levels of vocal synchrony in the first months of life predicted infant attachment security and maternal sensitivity, whereas low or high levels of synchrony did not (Hane, Feldstein, & Dernetz, 2003; Jaffe, Beebe, Feldstein, Crown, & Jasnow, 2001). A linear relation emerged between face-to-face synchrony and children's better cognitive outcomes (Feldman, Greenbaum, Yirmiya, & Mayes, 1996; Kirsh, Crnic, & Greenberg, 1995; Murray, FioriCowley, Hooper, & Cooper, 1996), higher symbolic competence (Feldman & Greenbaum, 1997), better self-regulatory skills (Feldman, Greenbaum & Yirmiya, 1999), and better social-emotional adaptation (Feldman & Eidelman, 2005; Harrist, Pettit, Dodge, & Bates, 1994), pointing to the important role of early affective matching in the normal development of child competencies. Finally, in the one available study involving children with autism, Siller and Sigman (2002) reported that when parents exhibited higher levels of synchronization toward their child with autism, the child showed better developmental outcomes (joint attention and language) years later.

The still-face paradigm comprises an extreme case of temporary social un-relatedness in which the mother refrains from any communicative acts after setting the stage for social interaction. Typically developing 3-month-old infants respond to their mothers' still face with gaze aversion, higher rates of scanning her face, reduced rates of positive affect, increased negative affect, and increased heart rate that signals their distress – that is, 'the still-face effect' (Kogan & Carter, 1996; Stoller & Field, 1982; Toda & Fogel, 1993; Tronick, Als, Adamson, Wise, & Brazelton, 1978; Weinberg & Tronick, 1996). The 'reunion effect', when the mothers resume interaction in the reunion play episode, is characterized by a mixed pattern of carryover negative affect and gaze aversion from the still-face episode (Toda & Fogel, 1993; Tronick, 1989; Tronick et al., 1978) and a rebound of positive affect. During the reunion episode, infants show more greeting behaviors, positive expressions, and positive affect compared to the first play and still-face episodes (Gusella, Muir, & Tronick, 1988; Weinberg & Tronick, 1996). Several still-face studies have been conducted with at-risk samples, including infants with Down syndrome (Berger & Cunningham, 1986; Carvajal & Iglesias, 1997; Legerstee & Bowman, 1989), in-uterus exposure to cocaine (Bendersky & Lewis, 1998), prematurity (Gutbrod, St. John, Rust, & Wolke, 2000; Segal et al., 1995), or deafness (Koester, 1995; Koester & Meadow-Orlans, 1999). Each of these infant samples differed from their typically developing peers in their responses to the still-face paradigm. In addition to these two well-established procedures, we developed a name-calling procedure in line with Baraneck's (1999), Osterling and Dawson's (1994), and Osterling, Dawson, and Munson's (2002) findings that children who were later diagnosed with autism respond less than other children to their name being called.

At 14 months, we administered the Early Social Communication Scales (Mundy, Hogan, & Doehring, 1996; Seibert, Hogan, & Mundy, 1982) to assess nonverbal communication skills. Communication skills comprise a focus of interest given the well-documented impairment that children with autism reveal in these skills. Such children show a specific and profound deficit in joint attention (Baron-Cohen, 1989; Hobson, 1989; Loveland & Landry, 1986; Mundy, Sigman, Ungerer, & Sherman, 1986), which involves coordination of attention between social partners (Scaife & Bruner, 1975), or what Trevarthen (1980) called shared subjectivity. Children with autism also show some difficulties in nonverbal requesting behaviors, but the more severe deficits in joint attention are linked to later theory of mind difficulties (Baron-Cohen, 1995; Charman et al., 2000) and predict language, social, and cognitive status years later (Sigman & Ruskin, 1999). The Checklist for Autism in Toddlers (CHAT; Baron-Cohen, Allen, & Gillberg, 1992; Baron-Cohen et al., 1996) was also employed because deficits in joint attention as assessed by the CHAT strongly predict later diagnoses of autism in undiagnosed toddlers at 18 months of age. In addition, the Bayley Scales of Infant Development – 2nd edition (BSID-II; Bayley, 1993) were employed at 4 and 14 months to assess general development and language. Whereas at 4 months, the kinds of social behaviors that could be measured are thought of as *precursors* to communication, at 14 months of age, possible differences in communication abilities may be assessed directly. To our knowledge, this study is the first to investigate longitudinally the development of young SIBS-A at the ages of 4 and 14 months. We predicted that at 4 months, infant SIBS-A would reveal less synchrony, be less affected by the still-face procedure, and respond less well to their name being called. At 14 months, we predicted that SIBS-A would display deficits in joint attention and communication as well as in general cognitive abilities as assessed by the Bayley Scales. Furthermore, we investigated the continuity between the 4 and 14 months measures.

Method

Participants

At age 4 months, the autism group comprised 21 dyads of mothers and their 4-month-old infants (8 girls, 13 boys) to whom an additional 10 dyads were added after age 4 months but before age 9 months. One sibling from the original SIBS-A group was seen at 18 months and thus excluded from data analyses at 14 months. At 14 months the autism group comprised 30 toddlers (11 girls/19 boys) who had an older sibling with autism. All probands were diagnosed with autism (for more information, please see the electronic appendix). Nine of the 31 probands with autism were classified as high-functioning (IQ and/or daily living skills scores ≥70) whereas the remaining 22 were classified as low-functioning (IQ and daily living skills scores <70). No significant differences emerged for any of the background independent measures within the group of SIBS-A based on probands' level of functioning.

The comparison group comprised 21 dyads of mothers and their infants (8 girls, 13 boys) who formed the SIBS-TD group at age 4 months, and 31 dyads (13 girls, 18 boys) at age 14 months. Families of the comparison group were recruited from maternity wards of the Hadassa Hospital in Mount Scopus, Jerusalem. Inclusion criteria required that the families were intact at the time of the study's initiation and that the older child exhibited typical development with no history of any learning and/or emotional difficulties and not receiving (nor had received in the past) any specialized interventions such as occupational therapy, speech and language therapy, or psychotherapy based on parental report. In addition, for both groups, inclusion criteria included participants' normal pregnancies as reported by the parents, with no peri-, pre-, or post-natal difficulties as well as normal gestational age (≥36 weeks).

At both ages, as best as possible, we matched the two groups on a one-to-one basis according to chronological age (CA), gender, birth order, number of children in the family, sex of the older proband and Bayley mental and motor scores. In addition, parents' age, ethnicity, income, and education level did not significantly differ between the two groups. Finally, the temperament profile (ICQ: Infant Characteristics Questionnaire; Bates, Freeland, & Lounsbury, 1979) revealed no significant differences between the two groups. Group characteristics at age 4 and 14 months are presented in Table 1.

The infants were seen again at ages 24 months, 36 months, and 54 months, employing various procedures that will be reported elsewhere. At all ages, a

Table 1 Sample characteristics for siblings of children with autism (SIBS-A) and siblings of children with typical development (SIBS-TD)

	4 months		14 months	
	SIBS-A $n = 21$	SIBS-TD $n = 21$	SIBS-A $n = 30$	SIBS-TD $n = 31$
Sibling's chronological age in weeks				
M	20.23	19.65	61.88	62.13
SD	3.24	2.83	1.26	1.26
Range	17–27	17–26	60–65	60–66
Temperament: Unpredictable				
M	31.43	28.71	32.33	33.59
SD	8.38	9.57	5.70	7.56
Range	18–50	14–48	18–43	19–49
Temperament: Fussy				
M	14.81	12.67	14.48	15.56
SD	3.81	4.19	3.29	3.39
Range	8–22	5–20	8–23	8–21
Temperament: Inadaptable				
M	10.33	11.05	12.11	12.93
SD	4.15	3.72	3.09	4.15
Range	5–20	5–19	6–18	6–24
Temperament: Dull				
M	12.90	12.76	10.82	10.52
SD	3.97	4.13	3.52	3.49
Range	8–22	5–20	5–19	5–19
Bayley mental score				
M	102.57	106.14	108.17	112.32
SD	7.85	6.28	15.38	7.58
Range	83–117	93–115	50–130	97–128
Bayley motor score				
M	99.38	101.23	105.80	108.97
SD	9.45	9.68	15.01	9.74
Range	86–116	86–121	50–124	84–124
Bayley Language Developmental age score (in months)*				
M			13.33	14.36
SD			2.38	.66
Range			9–17	13–15

Note. All comparisons with the exception of the Bayley Language Developmental Age Score at age 14 months are nonsignificant, p's > .01.
*$t_{(59)} = -2.30$, $p = .025$ (two-tailed, $d = .11$).

clinician trained in diagnosing autism spectrum disorders and difficulties participated in data collection. At age 14 months, one sibling was suspected of possibly having difficulties within the autism spectrum. The diagnosis of autism was confirmed at ages 24 and 36 months, using the ADI-R (Lord et al., 1994) and the Autism Diagnostic Observation Schedule – Generic (ADOS-G; Lord et al., 2000) conducted by two independent trained professionals. No other siblings showed any difficulties that necessitated an evaluation for autism spectrum conditions or for any other diagnostic entity. It should be noted that exclusion of the child who later developed autism from the analyses did not change the study outcomes (for a detailed description of this child's performance on the 4 and 14 months measures, please see the electronic appendix).

Measures administered at 4 and 14 months

Bayley Scales of Infant Development – 2nd edition (BSID-II; Bayley, 1993). The BSID-II is a standardized measure designed to assess the developmental level of infants and toddlers between the ages of 1 and 42 months. The Mental Developmental Index (MDI) and the Psychomotor Developmental Index (PDI) were calculated at 4 months and 14 months. An additional Language Developmental Age score was calculated at 14 months.

Infant Characteristics Questionnaire (ICQ; Bates, Freeland, & Lounsbury, 1979). The ICQ is a structured questionnaire designed to assess maternal perception of the infant's temperament. Mothers are requested to judge how well various behaviors describe their infant, on a scale ranging from *Not at all* (1) to *Very much* (8). The scores are summed to indicate four temperament characteristics: unpredictable, fussy–difficult, inadaptable, and dull. Each infant receives a score on each of the four temperament dimensions, resulting in four temperament scores for each infant.

Four months measures

Synchrony measured via mother-infant free play interaction. Mothers were instructed to play with the infant without toys, as they normally would at home, for 5 minutes. The interactions were coded using an adaptation of the Monadic Phase Manual (Tronick, Krafchuk, Ricks, Cohn, & Winn, 1985) to separately code maternal and infant states that incorporate affect and gaze information. Following Tronick et al. (1985), for each second, the mothers' and infants' behaviors were coded independently into five (for mothers) or six (for infants) exclusive categories or 'phases,' representing a continuum from negative to positive engagement. The mother's phases included: avert, object attend, social attend, object play, and social play. For infants, the additional phase of protest was included. Interrater reliability for the mother–infant interaction as assessed by Kappa coefficients was .85 for the mothers' behaviors and .80 for the infants' behaviors.

To score the existence or absence of significant infant–mother synchrony, each dyad received a score of 1 for the existence of a significant cross-correlation between the two time series (mother's and infant's) or a score of 0 for a nonsignificant cross-correlation. Dyads with significant synchrony were characterized as one of three types regarding dominance in leading the interaction: (1) baby leads, mother follows (BM); (2) mother leads, baby follows (MB); or (3) mutual synchrony, revealing both BM and MB synchrony types. In addition, a coherence score addressing the shared variance between the two time series after removing the autoregulated component was calculated, as well as a lag score indicating the time lag (in seconds) between a change in one partner's behavior and the corresponding change in the other partner's behavior. (For more information, please see the electronic appendix.)

Still-face paradigm. The procedure comprised three episodes. Mothers were instructed to move smoothly from one episode to the next when the experimenter softly knocked on the table. The first social play episode lasted 3–4 minutes (depending on how quickly the mother put on a still face following the signal after 3 minutes of play), followed by an expression of still face by the mother for 3 minutes, followed by resumed social interaction of reunion play lasting 3 minutes. Two coding procedures were utilized to code the still-face procedure: one for gaze and one for affect. The four independent raters who coded infant gaze were blind to participants' group affiliation and coded no other measures. Infant's gaze behavior was coded using an adaptation of the systems described by Mayes and Carter (1990), Carter, Mayes, and Pajer (1990), and Kogan and Carter (1996). Gaze in each second was coded as one of five exclusive gaze categories: gaze to mother's face; gaze to mother's hands and/or body; away/avert; gaze to object; and closed eyes. Kappa coefficients indicated excellent interrater reliability for infant gaze behaviors: .94 for the first play episode, .90 for the still-face episode, and .97 for the reunion play episode.

The additional four independent raters who coded infant facial affect were blind to participants' group affiliation and coded no other measures. Using an adaptation from Sigman, Kasari, Kwon, and Yirmiya (1992), the infant's facial affect expression was coded in 10-second intervals along the following 6-point scale: (1) very negative, (2) negative, (3) neutral, (4) interest, (5) positive, and (6) very positive. Thus, the interaction for each infant yielded about 6 observations of affect during the first play, 18 observations of affect during the still-face phase, and 6 observations of affect during the reunion play. Interrater reliability for the infant's affective expressions as assessed by Kappa coefficients was .93 during the first play episode, .89 during the still-face episode, and .89 during the reunion play episode.

For each of the five categories of infant gaze behavior during the still-face procedure, measures of percent duration were computed, representing the percent of the total time of each gaze category during the first play, still face, and reunion play episodes. The six infant facial affect categories during each of the three episodes were aggregated to form three mutually exclusive categories: negative affect (very negative and negative), neutral (neutral and interest), and positive (very positive and positive). Percentage scores were calculated for each category in each of the three episodes of the still-face procedure. (For more information, see the electronic appendix.)

Name-calling responsiveness. This new procedure examining responsiveness to name calling also followed a break (either after free play or after the still-face procedure). Infants were seated in an infant seat on a table, with a novel mobile placed in front and above the infant's head. Mothers initially stood about 60 cm from the infant seat on either the left or the right side. Mothers were instructed to begin calling out the infant's name once the infant engaged in looking at the mobile. Mothers called the infant's name for three times from each side for a total of 12 times, counterbalanced for initial side among participants. No physical contact or any other attention-getting procedures or gestures (e.g., clapping) were allowed. This interaction was videotaped with one camera focusing on the infant but with mother in view. One dyad from the SIBS-TD group did not participate in the procedure due to the infant's fussing, crying, and ensuing termination of the visit.

Two independent raters, who did not code any of the other measures, coded the name-calling procedure. For each of the name-calling attempts, the rater coded the infant's behavior as one of the following three responses: the infant neither searched nor found the mother, the infant searched but did not find the mother; or the infant searched and found the mother. Interrater reliability (Kappa coefficient) was .86. The first 3 name-calling sets, totaling 9 name-calling attempts, were included in the final analyses. Three scores were calculated for each of the two groups: the number of infants who did not search at all, who searched but did not find the mother, and who found the mother.

Fourteen months measures

Early Social Communication Scales (ESCS). The revised ESCS (Mundy et al., 1996; Seibert et al., 1982) provided data on the toddlers' nonverbal communication skills. This 20-minute structured assessment measured nonverbal communication skills such as joint attention behaviors, social play behaviors, and requesting behaviors between the ages of 6 and 30 months. For more information abut the procedure, please see Mundy et al. (1996). Three raters coded the videotaped ESCS procedure, computing the frequencies for each of the seven subscales: Initiating Social Interaction (ISI), Responding to Social Interaction (RSI), High and Low level of Initiating Joint Attention (HighIJA, LowIJA), Responding to Joint Attention (RJA) and High and Low level of Behavior Regulation (HighIBR, LowIBR). The interrater reliability for the seven ESCS scores based on 34% of the ESCS data resulted in $r = .94, .98, .88, .91, .87, .90, \& .90$, $p < .005$, for the total frequency scores of the ISI, RSI, HighIJA, LowIJA, RJA, HighIBR, and LowIBR, respectively. (For more information, please see the electronic appendix.)

Checklist for Autism in Toddlers (CHAT). The CHAT (Baron-Cohen et al., 1992) consists of two sections, one including 9 questions for the parent, and the other including 5 items to be completed by the experimenter based on observations. The 14 items on the scale yielded five key items, which are summed to yield a score ranging from 0 to 5 for each child. (For more information, see Baron-Cohen et al., 1992.)

Results

Preliminary *t*-test and χ^2 analyses did not reveal any significant sex differences on any of the independent or dependent measures at 4 or 14 months. Thus, further analyses were conducted independent of sex.

Four months

Mother–infant synchrony during free play interaction. Chi-square and independent *t*-test intergroup analyses were conducted to explore possible differences on mother–infant synchrony. Overall, significant synchrony of all types existed in 61.9% of the SIBS-A dyads and in 66.7% of the SIBS-TD dyads. Analyses did not reveal significant differences between groups in synchrony type or the two types of synchrony lag (see Table 2). However, examination of the coherence between mother's and infant's time series indicated that coherence was significantly lower in the SIBS-A group ($M = .13$, $SD = .06$) than in the SIBS-TD group ($M = .18$, $SD = .07$) when the interaction was led by the infant; $t_{(40)} = -2.316$, $p = .026$ (two-tailed, $d = .118$).

To further explore this finding, based on Jaffe et al. (2001), the groups were divided according to whether the dyads revealed low, mid-range, or high levels of coherence when the interaction was led by

Table 2 Group differences between siblings of children with autism (SIBS-A) and siblings of children with typical development (SIBS-TD) in synchrony during the free play interaction

Synchrony	SIBS-A $n = 21$	SIBS-TD $n = 21$	Group comparison
BM synchrony, N (%)	8 (38.1)	12 (57.1)	$\chi^2_{(1)} = 1.53$, $p = .22$
MB synchrony, N (%)	6 (28.6)	5 (23.8)	$\chi^2_{(1)} = .12$, $p = .73$
Mutual synchrony, N (%)	1 (4.8)	3 (14.3)	$\chi^2_{(1)} = 1.11$, $p = .29$
BM synchrony lag (in seconds), M (SD)	1.05 (1.53)	1.14 (1.56)	$t_{(40)} = -.20$, $p = .84$, d = .001
MB synchrony lag (in seconds), M (SD)	1.14 (2.20)	.62 (1.43)	$t_{(40)} = .92$, $p = .37$, d = .02
BM synchrony coherence, M (SD)	.13 (.06)	.18 (.07)	$t_{(40)} = -2.32$, $p = .03$, d = .12
MB synchrony coherence, M (SD)	.12 (.06)	.12 (.06)	$t_{(40)} = .02$, $p = .99$, d = .00

Note. BM = baby leads, mother follows; MB = mother leads, baby follows.

the infant. These coherence categories were operationalized utilizing the mean of all participants (SIBS-A and SIBS-TD), which was .154 with a standard deviation of .07. Thus, synchrony below the middle range was defined as a score lower than .083 (1 *SD* below the mean), mid-range synchrony was defined as a score of .083 to .224 (within the range of 1 *SD* below or above the mean), and synchrony above the middle range was defined as a score higher than .224 (1 *SD* above the mean). Among the SIBS-A group, 5 dyads received synchrony scores below mid-range, 14 within mid-range, and 2 above mid-range. Within the group of SIBS-TD, 0 received synchrony scores below mid-range, 16 within mid-range, and 5 above mid-range. A likelihood ratio test revealed a significant difference between the groups (of 8.39, df = 2, $p < .01$). Thus, both intergroup analyses of mean differences and analysis regarding the number of dyads within each group revealed that SIBS-A dyads were less synchronous compared to SIBS-TD dyads.

Still-face procedure. A significant difference emerged between the two groups in the duration of the still-face episode. Duration was longer for SIBS-A ($M = 110.06$ seconds, $SD = 48.74$) than for SIBS-TD ($M = 75.47$ seconds, $SD = 45.46$), $t_{(35)} = 2.233$, $p = .032$ (two-tailed, $d = .125$). Therefore, analyses regarding infant gaze and affect were carried out using percentage scores. Further examination revealed that 8 of the 19 infants in the SIBS-TD group started to fret or cry which resulted in their mothers stopping the still-face episode, whereas only 2 of the 18 infants in the clinical group did so (Z for comparison between proportions = 2.12, $p < .05$), suggesting that SIBS-A were less upset by the procedure compared to SIBS-TD.

Infant gaze. In Table 3, descriptive data including percentage means and standard deviations for infant gaze during the still-face procedure are presented. To explore possible group differences, a separate 2-way analysis of variance (ANOVA) with repeated measures for the three episodes of the still-face procedure (first play, still-face, and reunion play) was conducted for each of the five gaze categories. The ANOVAs all yielded nonsignificant group and interaction effects; thus, the gaze behavior of SIBS-A did not differ from the gaze behavior of SIBS-TD.

Infant affect. Descriptive data including percentage means and standard deviations are presented in Table 3. A separate 2-way ANOVA with repeated measures for the three episodes of the still-face procedure was conducted for each of the three aggregated facial affect categories. A group effect emerged only for the neutral affect category, in that SIBS-A displayed significantly more neutral affect throughout the still-face procedure ($M = 79.85\%$, $SD = 18.79$) than did SIBS-TD ($M = 67.89\%$, $SD = 23.72$), $t_{(33)} = 2.061$, $p = .047$, two-tailed, $d = .11$). All other group and interaction effects were nonsignificant.

Still-face and reunion effects. SIBS-A did not differ in their socio-emotional reactions to the still-face perturbation compared to infant SIBS-TD, and revealed the well-documented patterns of still-face and reunion effects. From the first play episode to the still-face episode, both groups revealed a significant decrease in positive affect and in gaze toward the mother's hands and/or body as well as a significant increase in averted gaze, closed eyes, and negative affect behaviors. The 'reunion effect' was manifested in both groups by a continued higher level of closed eyes and negative affect during the reunion play episode; these carryover effects from the still-face episode surpassed those behaviors exhibited in the first play episode. In addition, a significant rebound of positive affect emerged when mothers resumed interaction in the reunion play episode.

Name-calling procedure. Data were analyzed using z tests for proportions. Surprisingly, the proportion of infants who responded to their name being called and who searched and found their mother at least once was significantly higher in the SIBS-A group (17 of 21) than in the SIBS-TD group (6 of 20), $z = 3.276$,

Table 3 Infant Gaze and Affect Data during the Still-Face Procedure: Means and Standard deviations

		SIBS-A(n = 18)			SIBS-TD (n = 19)		
		First Play %	Still Face %	Reunion Play %	First Play %	Still Face %	Reunion Play %
Gaze							
Toward mother's face	*M (SD)*	45.71 (32.75)	31.31 (25.36)	40.46 (32.92)	44.58 (24.27)	47.72 (30.05)	35.36 (32.31)
Toward mother's body	*M (SD)*	19.77 (21.59)	8.41 (15.83)	21.24 (21.24)	24.25 (21.73)	1.97 (5.01)	12.74 (16.39)
Away/Avert	*M (SD)*	24.80 (22.23)	58.60 (22.77)	27.60 (27.62)	22.03 (26.51)	43.56 (30.11)	31.94 (31.87)
Toward object	*M (SD)*	9.72 (18.92)	.49 (2.10)	2.26 (6.07)	8.46 (18.45)	0.00 (00)	11.50 (24.99)
Closed eyes	*M (SD)*	0.00 (00)	1.21 (2.41)	8.44 (24.33)	0.68 (1.73)	6.75 (12.80)	8.47 (23.12)
Affect							
Negative	*M (SD)*	3.53 (10.57)	20.14 (29.89)	20.74 (30.75)	12.59 (22.48)	41.69 (43.66)	28.33 (35.57)
Neutral	*M (SD)*	88.43 (16.38)	78.75 (29.47)	73.70 (29.83)	78.15 (23.66)	57.83 (43.44)	62.02 (32.56)
Positive	*M (SD)*	8.04 (14.72)	1.11 (4.71)	5.56 (11.07)	9.26 (16.39)	.48 (2.09)	9.65 (17.63)

$p < .05$. No significant group difference emerged for the proportion of infants who responded and searched at least once but did not find the mother, $z = 1.105$, $p > .05$ (SIBS-A: 1 of 21; SIBS-TD: 3 of 20). The proportion of infants who did not respond to their name being called and who neither searched nor found the mother at all was significantly higher in the SIBS-TD group (11 of 20) than in the SIBS-A group (3 of 21), $z = 2.75$, $p < .05$.

To ensure that no procedural bias led to the finding whereby more SIBS-A than SIBS-TD responded to their name being called and searched and found their mother, the videotaped name-calling procedure was re-coded for (a) duration of the first 3 trials, (b) number of times that the mother called her infant's name during the first 3 trials, and (c) number of times per minute that the mother called her infant's name throughout 30 minutes of the session (excluding the name-calling procedure). No significant differences emerged for any of these measures (all $p > .05$, $d \leq .09$). Full data are available from the first author.

Fourteen months

Intergroup analyses.Toddlers' developmental level (BSID-II). As can be seen in Table 1, the two groups did not significantly differ on their overall Bayley mental score or on their overall Bayley motor score. However, significant intergroup differences did emerge on the Bayley language score, with a significantly lower developmental language age among SIBS-A than among SIBS-TD. A closer examination of the data revealed that the language developmental age of 8 SIBS-A was lower than 14 months: Six toddlers achieved a language developmental age of 9 months, and 2 toddlers achieved a language developmental age of 12 and 13 months respectively. Only 3 SIBS-TD achieved a language developmental age below 14 months and all 3 had a delay of 1 month only (language developmental age of 13 months).

Given these findings, we reanalyzed the Bayley scores using ANOVA with three groups; the 6 SIBS-A with substantial language delay of 5 months (SIBS-A-LD), the remaining 24 SIBS-A with normal language levels (SIBS-A-nonLD), and the 31 SIBS-TD.

These analyses revealed that, as expected, the 6 SIBS-A-LD had a significantly lower Bayley language age score ($M = 9$, $SD = 0$) compared to the 24 SIBS-A-nonLD ($M = 14.42$, $SD = 1.02$) and to the 31 SIBS-TD ($M = 14.35$, $SD = .66$), who did not differ from each other ($F_{(2,58)} = 123.08$, $p < .001$ followed by Scheffé analyses, $p < .001$). Similar results emerged for the Bayley mental score in that the 6 SIBS-A-LD had a significantly lower mental score ($M = 91$, $SD = 21.33$) compared to the 24 SIBS-A-nonLD ($M = 112.46$, $SD = 10.16$) and to the 31 SIBS-TD ($M = 112.32$, $SD = 7.58$), who did not differ from each other ($F_{(2,58)} = 11.26$, $p < .001$ followed by Scheffé analyses, $p < .001$). A trend was found between the 3 groups on the Bayley motor score ($F_{(2,58)} = 2.50$, $p = .09$), with the 6 SIBS-A-LD achieving a lower Bayley motor score ($M = 96.83$, $SD = 24.89$) compared to the 24 SIBS-A-nonLD ($M = 108.04$, $SD = 11.10$) and to the 31 SIBS-TD ($M = 108.97$, $SD = 9.74$).

Nonverbal communication (ESCS). We conducted a MANOVA followed by *t*-test analyses to examine possible group differences on the ESCS measures. No significant overall group effect emerged for the total of the seven ESCS variables, $F_{(7,53)} = 1.77$, $p = .113$. However, inasmuch as this study constituted the first that investigated ESCS with young SIBS-A, we continued by examining possible intergroup differences on each of the seven ESCS categories. These analyses revealed one significant difference in the ESCS category of High-level requesting behaviors (HighIBR): SIBS-A initiated fewer high-level requesting behaviors ($M = 5.32$, $SD = 4.12$) than did SIBS-TD ($M = 9.48$, $SD = 6.14$), $F_{(1,59)} = 9.6$, $p = .003$, (two-tailed, $d = .14$). All six other comparisons between the two groups revealed nonsignificant differences (see Table 4).

We reanalyzed the data using ANOVA with 3 groups. These analyses revealed significant findings for Low and High levels of initiating requesting behaviors. Regarding initiations of High levels of requesting behaviors, the 6 SIBS-A-LD ($M = 4.67$, $SD = 5.43$) did not differ significantly from the 24 SIBS-A-nonLD ($M = 5.48$, $SD = 3.85$) and nor from the SIBS-TD ($M = 9.48$, $SD = 6.14$), yet the 24 SIBS-A-nonLD revealed significantly fewer High-level

Table 4 Group differences between siblings of children with autism (SIBS-A) and siblings of children with typical development (SIBS-TD) during the ESCS: means and standard deviations

Category	SIBS-A $n = 21$	SIBS-TD $n = 21$	Group comparison
Initiates Social Interaction (ISI), M (SD)	1.57 (1.36)	2.03 (1.11)	$t(59) = -1.47$, $p = .15$, d = .04
Responds to Social Interaction (RSI), M (SD)	12.27 (5.60)	13.56 (4.83)	$t(59) = -.97$, $p = .34$, d = .02
Initiates Joint Attention – Low (LowIJA), M (SD)	4.77 (5.36)	4.42 (4.65)	$t(59) = .27$, $p = .79$, d = .001
Initiates Joint Attention – High (HighIJA), M (SD)	1.10 (1.97)	1.61 (2.28)	$t(59) = -.94$, $p = .35$, d = .02
Responds to Joint Attention (RJA), M (SD)	3.08 (1.92)	3.65 (1.32)	$t(59) = -1.36$, $p = .18$, d = .03
Initiates Behavior Requesting – Low (LowIBR), M (SD)	20.71 (11.47)	25.76 (11.76)	$t(59) = -1.69$, $p = .10$, d = .05
Initiates Behavior Requesting – High (HighIBR), M (SD)	5.32 (4.12)	9.48 (6.14)	$t(59) = -3.10$, $p = .003$, d = .14

requesting behaviors compared to SIBS-TD ($F_{(2,58)} = 4.79$, $p < .05$ followed by Scheffé analyses, $p < .05$). Regarding initiating Low levels of requesting behaviors, the 6 SIBS-A-LD ($M = 12.83$, $SD = 11.37$) did not differ significantly from the 24 SIBS-A-nonLD ($M = 22.68$, $SD = 10.85$), who did not differ from the SIBS-TD ($M = 25.75$, $SD = 11.76$), yet the 6 SIBS-A-LD revealed significantly fewer Low-level requesting behaviors compared to SIBS-TD ($F_{(2,58)} = 3.30$, $p < .05$ followed by Scheffé analyses, $p < .05$).

CHAT. On the 5 key item score, 3 SIBS-A received a score of 3, whereas none of the SIBS-TD did so. An additional 4 SIBS-A received a score of 2, compared to 6 SIBS-TD. Seventeen SIBS-A and 15 SIBS-TD received a score of 1. We analyzed the CHAT data using the 3 groups, SIBS-A-LD, SIBS-A-nonLD and SIBS-TD. These analyses revealed that the 6 SIBS-A-LD had a significantly higher CHAT score ($M = 1$, $SD = 1.10$) compared to the 24 SIBS-A-nonLD ($M = .17$, $SD = .38$) and to the 31 SIBS-TD ($M = .13$, $SD = .34$), who did not differ from each other ($F_{(2,58)} = 8.98$, $p < .001$ followed by Scheffé analyses, $p < .01$).

Continuity between 4 and 14 months

We examined the continuity of our findings for the 20 SIBS-A and 21 SIBS-TD who participated at both ages. First, at 4 months, compared to SIBS-TD, SIBS-A revealed less synchrony with the mother when the infant led the mother–infant interaction. Associations were nonsignificant between the above synchrony measure and all of the 14-month measures of cognition, language and nonverbal communication for both group. Thus, we found no evidence for continuity between our synchrony measures at age 4 months and our measures at 14 months for this small sample.

Second, at age 4 months SIBS-A displayed more neutral affect compared to SIBS-TD during the still-face procedure. Within the group of SIBS-A, correlational analyses revealed that the percentage score of neutral affect during the still-face procedure was significantly negatively correlated with the frequency of initiating high-level behaviors of joint attention (High IJA) ($r = -.56$, $p = .009$). Thus, SIBS-A who showed more neutral affect during the still-face procedure at age 4 months initiated less joint attention at the age of 14 months. This association was nonsignificant for the SIBS-TD group. Furthermore, in both groups of siblings, neutral affect display during the still-face procedure at age 4 months did not correlate significantly with any of the Bayley or CHAT measures at age 14 months.

Third, the 4-month data indicated that more SIBS-A responded to their name being called and searched and found their mothers compared to SIBS-TD. Thus, we next explored whether those infants within each group who responded and searched and found the mother, as compared to those who did not respond, differed on any of the 14-month measures. Within the SIBS-A group, toddlers who did not respond nor found their mothers at age 4 months during the name-calling procedure ($n = 4$) initiated fewer requesting acts (High IBR) at the age of 14 months ($M = 2.25$, $SD = 1.71$) than did toddlers who had responded and searched and found their mothers ($n = 16$, $M = 6.06$, $SD = 4.42$), $t_{(18)} = -2.73$, $p = .008$. None of the other analyses within the SIBS-A group was significant, employing the other ESCS variables, the Bayley scores (mental, motor, language), or the CHAT score at age 14 months. Regarding the SIBS-TD group, nonsignificant differences emerged on all the dependent variables between those infants who responded and searched and found their mother and those who did not.

Next, given the identification of the 6 SIBS-A–LD at age 14 months, we reanalyzed the 4 months measures that were used to match the samples on (i.e., Bayley scores, temperament, demographic information), employing ANOVAs for 3 groups (SIBS-A-LD, SIBS-A-nonLD and SIBS-TD). All analyses were nonsignificant. Examination of the dependent measures revealed a significant difference among the three groups for the synchrony variable in the baby lead, mother follows dyads ($F_{(2,39)} = 3.52$, $p < .05$). The 5 SIBS-A-LD were less synchronous ($M = .10$, $SD = .06$) compared to the 16 SIBS-A-nonLD ($M = .14$, $SD = .06$) and to the 21 SIBS-TD ($M = .18$, $SD = .07$). However, Scheffé post-hoc comparisons revealed that only the difference between SIBS-A-LD and SIBS-TD approached significance ($p = .06$). For the still-face procedure, the ANOVA revealed a trend ($F_{(2,34)} = 2.86$, $p = .07$), with the 5 SIBS-A-LD showing more neutral affect ($M = 90.44\%$, $SD = 12.11$) compared to the 13 SIBS-A-nonLD ($M = 76.37\%$, $SD = 19.17$) and to the 18 SIBS-TD ($M = 66.21\%$, $SD = 23.76$). However, post-hoc Scheffé comparisons were nonsignificant (all p's $>.05$). On the name-calling procedure, no differences emerged between the SIBS-A-LD and SIBS-A-nonLD (all 5 SIBS-A-LD searched for their mother and 4 of the 5 found her; 13 of the 16 SIBS-A-nonLD searched and found).

Discussion

Siblings of children with autism and SIBS-TD were examined at the ages of 4 and 14 months using various measures assessing social/emotional development, communication and cognition. During a free-play, face-to face interaction between the mothers and infants at age 4 months, SIBS-A dyads were less synchronous during the interactions led by the infant. During the still-face procedure, SIBS-A were less upset by it and revealed more neutral affect. Third, unexpectedly, more SIBS-A responded to their name being called and searched and found their mother compared to SIBS-TD. At 14 months, SIBS-A initiated fewer nonverbal requesting gestures and achieved as a group lower language scores on the Bayley Scales. Six SIBS-A revealed a language delay of 5 months and were responsible for the differences found between SIBS-A and SIBS-TD on the Bayley language score. In addition, the Bayley mental score of SIBS-A-LD was significantly lower than that of SIBS-A-nonLD and SIBS-TD and their CHAT score was significantly higher.

In the face-to-face free play interaction at the age of 4 months, the present findings regarding synchrony resemble those reported for typically developing infants (Feldman, 2003; Lester et al., 1985; Tronick & Cohn, 1989). Synchronies of all types were found in more than 60% of the dyads in both groups, with infants more dominant in leading the interaction. However, synchrony was stronger in dyads of the SIBS-TD group during infant-led interactions. Furthermore, the distribution of low, mid-range, and high levels of this type of synchrony differed between the two groups, with more SIBS-A manifesting the less optimal levels of synchrony. Indeed, not one SIBS-TD dyad exhibited low synchrony, whereas 5 SIBS-A dyads (24%) did. These findings indicate a closer affective match between mothers and their infants in the group of SIBS-TD in infant-led interactions. Our finding from the three-group analysis (SIBS-A, SIBS-A-nonLD, SIBS-TD) may suggest that mothers may have been picking up on some characteristics of their babies since synchrony was weaker primarily in the infants who later were to have language delays. However, this difference may be due to a lack of experience among mothers of SIBS-A in reacting to their infant's initiative expressions, which may have been absent in previous interactions with the older child who was later diagnosed with autism. This difference may also stem from greater maternal stress or other maternal characteristics, which were not investigated in the current study and which merit further research in the future.

Infant SIBS-TD were more sensitive to the still-face episode and more upset by it, as indicated by the higher number of infants in this group who started to fuss and cry, which resulted in the mother's cessation of her still-face behavior. Furthermore, the SIBS-A significantly lower level of reactivity to the still-face procedure was also evident in the overall higher duration of neutral affect displayed by SIBS-A compared to SIBS-TD throughout the still-face procedure. However, as found in normative studies (Fogel, 1993; Toda & Fogel, 1993; Weinberg & Tronick, 1996), the two groups of siblings displayed the same signs of distress and disengagement that signify the still-face effect: significant increases in gaze aversion, closing of the eyes, and negative affect, as well as significant decreases in gaze toward the mother's hands and/or body, gaze toward an object, and positive affect, from the first play episode to the still-face episode. These findings suggest that although SIBS-A at the age of 4 months are somewhat less affected by the procedure and reveal more neutral affect than their SIBS-TD peers, the family history of autism does not disrupt these infants' demonstration of the documented still-face and reunion effects. Thus, at the age of 4 months, the SIBS-A group and the SIBS-TD group did not differ significantly on most of the early social engagement measures, indicating that infant SIBS-A are functioning well at age 4 months and that we were unable to identify early markers for later difficulties at this age with the measures employed in the current study.

On the name-calling procedure, significantly more SIBS-A responded to their name being called and searched and found the mother compared to SIBS-TD. This finding was surprising given previous reports that identified a lack of response to name calling as a chief characteristic of young infants who were later diagnosed with autism (Osterling & Dawson, 1994; Osterling et al., 2002). The present findings that SIBS-A were less reactive and more emotionally neutral during the still-face procedure but more responsive during the name-calling procedure may seem contradictory. However, it is important to note that the free play face-to-face interaction and the still-face procedures are inherently less structured and goal-oriented compared to the name-calling procedure. The former two procedures thus allow more flexibility for both partners, whereas the latter procedure is more structured by one partner (the mother) and has the definite goal of obtaining the infant's attention. Perhaps these mothers, who already have an older child with autism in the family, initiate more interactions with their infants to assure themselves that their young infants are developing well. It will be important to try to replicate this procedure in future studies.

At the age of 14 months, the full sample of SIBS-A revealed a significant delay in language development, and produced significantly fewer requesting behaviors such as giving and pointing with or without eye contact, as assessed by the ESCS. Our longitudinal analyses revealed that within the group of SIBS-A, those siblings who had displayed more neutral affect during the still-face procedure at age 4 months initiated fewer joint attention behaviors at age 14 months. Also, those SIBS-A who had not searched for the mother in the name-calling procedure at age 4 months later, at 14 months, initiated fewer behavior regulation acts.

Once the group of SIBS-A was divided into 2 groups, one with and one without a language delay, reanalyses of the 14 months data confirmed that at the current testing time and as evaluated by the current measures, most SIBS-A are functioning well and that only a few have difficulties and delays. These findings suggest that at age 14 months, some SIBS-A (about 20%) already show evidence of some subtle deficits, which appear in some of the same domains most severely affected in autism: Communication and language.

However, whereas impairments in joint attention comprise the most striking nonverbal communications skills deficit in children with autism (Baron-Cohen, 1989; Charman et al., 1997; Mundy et al., 1986), the younger siblings of these children with autism showed impairments in skills involving behavior regulation such as reaching and giving.

Several possible explanations exist for the finding that SIBS-A display difficulties in initiating requesting behavior and language. First, studies of the 'broad phenotype' in other first-degree relatives of children with autism, such as parents, have reported both social and language abnormalities (Baron-Cohen & Hammer, 1997; Bolton et al., 1994; Landa, Piven, Wzorek, Gayle, & Folstein, 1992), implying that the demonstrated deficits in SIBS-A may have a genetic basis. The broad phenotype may be evident because possession of some but not all of the susceptibility genes for autism may be expressed as mild symptoms or partial syndromes (Bailey et al., 1995; Piven, Palmer, Jacobi, Childress, & Arndt, 1997). SIBS-A and other relatives may be expected to express other aspects of the broad phenotype not assessed in the present study, such as unusually good attention to detail (Baron-Cohen & Hammer, 1997; Briskman, Happé, & Frith, 2001) and talent in skills related to systemizing (Baron-Cohen, 2002). Such cognitive assets, and not just areas of deficit, should be assessed in future studies of SIBS-A.

An alternative explanation for the present results concerning the SIBS-A group's delayed language and reduced requesting behavior may pinpoint such siblings' learning environment and the special family environment in which they grow up. It might be argued that fewer opportunities exist for communication with their sibling with autism, and that parents disproportionately divert their attention to the sibling with autism, thus decreasing parents' availability as communication partners. Whilst research has shown demonstrable effects of having a sibling with autism on the behavior of the siblings without autism (Howlin, 1987), it is not clear that these specific results would have been predicted from such an environmental theory. When we initiated our study, to the best of our knowledge, it was the first to investigate the social engagement, communication, and cognitive development of young SIBS-A from 4 to 14 months employing interactional procedures.

As a first step, it suffers from some shortcomings, such as the relatively small sample size, lack of clinical information regarding the mothers (i.e., stress levels, anxiety), and the lack of additional comparison groups, such as a group of siblings of children with learning disorders or mental retardation.

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Supplementary Material

The following supplementary material is available for this article online:

Details of empirical analysis: Participants, procedure and results. This material is available as part of the online article from http://www.blackwell-synergy.com