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ORIGINAL PAPER

Early Social and Emotional Communication in the Infant Siblings of Children with Autism Spectrum Disorders: An Examination of the Broad Phenotype

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Abstract Infants with older siblings with Autism Spectrum Disorders (ASD-sibs) are at risk for socio-emotional difficulties. ASD-sibs were compared to infants with typically developing older siblings (TD-sibs) using the face-to-face/still-face (FFSF) at 6 months and the Early Social Communication Scale (ESCS) at 8, 10, 12, 15, and/or 18 months. ASD-sibs smiled for a lower proportion of the FFSF than TD-sibs and lacked emotional continuity between episodes. With respect to TD-sibs, ASD-sibs engaged in lower rates of initiating joint attention at 15 months, lower rates of higher-level behavioral requests at 12 months, and responded to fewer joint attention bids at 18 months. The results suggest subtle, inconsistent, but multi-faceted deficits in emotional expression and referential communication in infants at-risk for ASDs.

Keywords Autism spectrum disorders · Siblings · At-risk · Emotion · Facial expressions · Joint attention

Introduction

Autism Spectrum Disorders (ASD) are pervasive developmental disorders adversely affecting the social, cognitive, and frequently, the intellectual functioning, of diagnosed individuals (Ritvo, Freeman, Pingree, & Mason-Brothers, 1989). There is evidence that a broad ASD phenotype characterizes relatives of individuals diagnosed with ASDs (Bailey, Palferman, Heavey, & Le Couteur, 1998; Constantino et al., 2006; Dawson et al., 2002). The purpose of this paper is to assist in the identification of early social and emotional deficits characteristic of the broad ASD phenotype by examining infant siblings of children with an ASD.

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The Broad ASD Phenotype

ASDs are genetically linked developmental disorders involving key deficits in communicating positive emotion, initiating joint attention (IJA), engaging in reciprocal interaction, and using language. Autism is characterized by difficulties in three separate areas: social and emotional functioning, communication, and sensory concerns or restricted or repetitive interests. Other disorders on the autism spectrum include most, but not all, of these deficits. The broad phenotype of autism involves more subtle ASD-linked deficits, such as language delays, difficulties with sensory integration, and potential difficulties with emotion regulation and communication.

ASDs present a broad range of manifestations, often within the same family. In identical twin pairs, one member of whom has autism, the concordance rate for autism is 60% and the concordance rate for a disorder on the autism spectrum is more than 90% (Veenstra, Vanderweele, & Cook, 2003). More than 10% of full siblings of probands with autism show deficits associated with an ASD diagnosis (Landa & Garrett Mayer, 2006; Zwaigenbaum et al., 2005). Among adult full siblings of probands with autism, as many as 20% show a deficit in social or communicative functioning as indicated by family history (Bolton, Macdonald, Pickles, & Rios, 1994; Pickles et al., 2000; Rutter, 2000). Deficits in social responsivity among sibling pairs illustrate the concept of the broad ASD phenotype (Constantino et al., 2006). Levels of social responsivity in twin-pairs are associated even when neither twin is diagnosed with an ASD (Constantino & Todd, 2003).

ASDs are typically not diagnosed until late toddlerhood or pre-school age. Parents, however, report that the mean age of ASD symptom recognition is between 16 and 20 months (Chakrabarti & Fombonne, 2005; Ozand, Al Odaib, Merza, & Al Harbi, 2003; Short & Schopler, 1988; Spitzer & Siegel, 1990; Volkmar, Stier, & Cohen, 1985) and many parents report developmental deficits within the first year of life (Zwaigenbaum et al., 2005). Little, however, is known about ASD-related deficits in infancy. ASDs occur in approximately one in every 150–250 pre-school children, making the disorder too rare to study prospectively in the general population (Bryson & Smith, 1998; Chakrabarti & Fombonne, 2001).

Retrospective studies of home videotapes suggest that deficits are present as early as 3–8 months of age in infants who are later diagnosed with autism (Adrien, Faure, Perrot, & Hameury, 1991; Werner, Dawson, Osterling, & Dinno, 2000; Werner & Dawson, 2005; Werner, Dawson, Munson, & Osterling, 2005). Parent report may be confounded by selective recall, at times lack of knowledge about development, and difficulties associated with admitting developmental difficulties in their child (Chawarska & Volkmar, 2003; Stone, Hoffman, Lewis, & Ousley, 1994), making prospective examination of the development of ASDs important. This is the rationale for the current study of the early autism phenotype in young siblings of children with ASD.

We investigated whether infant siblings of children with an ASD (ASD-sibs) showed developmental differences associated with the broad ASD phenotype between the ages of 6 and 18 months. We compared ASD-sibs to infants with typically developing siblings (TD-sibs) and focused on early measures of emotion expressed in parent–child interactions and later measures of joint attention.

Emotional Expression and the Broad ASD Phenotype

The first degree relatives of individuals with autism evidence an increased expression of personality characteristics such as social isolation (Bolton et al., 1994; Piven, Wzorek, Landa, & Lainhart, 1994; Wolff, Narayan, & Moyes, 1988) and social phobia (Smalley, McCracken, & Tanguay, 1995). They also show increased expression of personality traits such as anxiousness, impulsiveness, and irritableness (Murphy et al., 2000).

Retrospective studies of the early development of children with autism suggest that attenuation of positive affect, or of the tendency to socially direct positive affect, may be one of the earliest markers of atypical development (Baranek, 1999; Osterling, Dawson, & Munson, 2002; Wimpory, Hobson, Williams, & Nash, 2000). Parents of a child with an ASD perceive their child's emotional expressions to be more negative and less positive than parents of children who are mentally retarded (Capps, Kasari, Yirmiya, & Sigman, 1993).

The face-to-face/still-face (FFSF) is an interactive protocol useful for assessing the positive and negative emotional expressivity of infants (Adamson & Frick, 2003; Tronick, Adamson, Wise, & Brazelton, 1978). Parents are asked to play normally with their infant, hold a still face, and then resume play. Positive engagement declines and negative engagement increases when the parent ceases play and holds a still face (Tronick et al., 1978). The resumption of play in the reunion is characterized by moderate levels of both positive and negative emotional expressions (Moore, Cohn, & Campbell, 2001). In normative (Weinberg, Tronick, Cohn, & Olson, 1999) and at-risk (Acosta, Messinger, Cassel, & Bauer, 2004) samples, infants showed between-episode stability in emotional expressivity. Levels of negative affect are correlated between the face-to-face (FF), still face (SF), and reunion (RE) episodes, as are levels of positive affect.

Children with autism display flatter or more neutral affect than comparison children with mental retardation (Yirmiya, Kasari, Sigman, & Mundy, 1989). In a recent prospective study, 4-month-old ASD siblings showed more neutral affect than TD siblings in response to a maternal SF. Fewer ASD than TD siblings engaged in negative affect over the course of the SF, although mean differences in negative affect did not achieve significance (Yirmiya et al., 2006). In another prospective study, 12-month-old ASD siblings who later revealed autistic symptomatology showed deficits in social smiling, displaying less positive emotion than typically developing infant siblings (Zwaigenbaum et al., 2005). We expected ASD-sibs to display less positive and more negative emotion in the FFSF than TD-sibs at 6 months of age, but also expected they would have a propensity toward neutral, potentially disengaged, affect.

Referential Communication and the Broad ASD Phenotype

Referential communication involves conventional gestures used to attain objects and responses to and initiations of joint attention. The Early Social Communication Scales (ESCS) were developed to assess for these early communicative behaviors in young children who do not yet use language as their primary mode of communication (Mundy, Hogan, & Doehring, 1996; Mundy et al., in submission). IJA refers to the use of gestures and gaze to declaratively or proto-declaratively communicate about an object or event in the environment (Jones & Carr, 2004; Messinger & Fogel, 1998). IJA behaviors are precursors to language and may be especially important as predictors of later differences in social, cognitive, and behavioral outcomes. IJA behaviors at 18 months predict language development at 2 years of age (Mundy et al., in submission). Deficits in IJA (Baranek, 1999; Jones & Carr, 2004) and other broader social deficits (Werner et al., 2005) are common in children with an ASD, and are frequently evident in children with early onset (e.g., non-regressive) autism before 1 year of age. In fact, such deficits discriminate approximately 80–90% of children with autism from children with other developmental delays (Lewy & Dawson, 1992; Mundy, Sigman, Ungerer, & Sherman, 1986), but not until after the first birthday. In a recent study, ASD siblings (14–19 months of age) exhibited significantly less IJA than young TD children (10–19 months of age) (Goldberg et al., 2005).

Responding to joint attention (RJA) refers to the child's ability to follow the joint attention behavior (i.e., pointing) of the examiner. Associations have been found between RJA and later language development in infants between 6 and 18 months of age (Morales et al., 2000). Examinations of RJA in young ASD siblings have yielded mixed results (Goldberg et al., 2005; Presmanes, Walden, Stone, & Yoder, 2007; Yirmiya et al., 2006). While Yirmiya et al. (2006) found no RJA differences between groups, others have found ASD-sibs to show RJA decrements (Goldberg et al., 2005) and related deficits in responding to an adult's referencing behaviors (Presmanes, Walden, Stone, & Yoder, 2007).

Initiating behavioral requests (IBR) refers to requests for help or an object (Mundy et al., 1996). Lower level IBR behaviors include making eye contact in order to request a toy or reaching toward a toy. Higher level IBR behaviors include pointing at a desired toy or giving the examiner a toy. Yirmiya et al. (2006) found that ASD siblings engaged in fewer higher-level IBRs than TD-sibs while Goldberg et al. (2005) found ASD-sibs engaged in significantly less IBR of all types than young TD children. We expected ASD-sibs to display lower levels of IJA, RJA, and IBR than TD-sibs during the Early Social Communication Scales (ESCS) between 8 and 18 months of age.

Method

Participants

Infants with older siblings (N = 31) were assessed with respect to their social and emotional functioning in the first year and a half of life. Infants in this study were part of a larger sample investigating the social, emotional, and cognitive development of ASD-sibs and TD-sibs. Infants were included in this sample if they participated in a 6-month assessment and participated in at least one of the 8, 10, 12, 15, or 18-month assessments.

There were twelve ASD-sibs. Eleven infants had older full-siblings on the autism spectrum and one infant had a half-sibling on the autism spectrum (included in the ASD-sibs group). There were 19 TD-sibs. The status of the older sibling was obtained by parent report at the initial telephone contact. Infants were included in the ASD-sibs group if their older sibling was diagnosed with Autism, Asperger's Disorder, or Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS). We are currently in the process of confirming this information via independent assessments conducted at our laboratory. Currently, 15 TD-sibs are 18 months of age or older and six ASD-sibs are 18 months of age or older. The older typically developing siblings ranged in age from 2.22 to 15.18 years (M = 6.66, SD = 3.29) and the older siblings diagnosed with an ASD ranged in age from 3.56 to 11.27 (M = 7.25, SD = 2.55), a non-significant difference, p = .60. The TD-sibs were Hispanic (36.8%), Caucasian (47.4%), African American (5.3%), Asian (5.3%), and biracial (5.3%). The ASD-sibs were Hispanic (66.7%) and Caucasian (33.3%).

Procedure

All infants participated with a parent in the FFSF protocol (Tronick, Als, & Brazelton, 1977) at a mean age of 6.15 months (SD = .33; range 5.03–6.87 months). There was no age of FFSF administration difference between groups, $p > .50$. In the FFSF parents were asked to play with their baby without toys for 3 minutes (FF), stop playing and maintain a still face with no emotional expression for 2 minutes (SF), and resume play for another 3 minutes (RE). Infants were placed in a car seat during the FFSF and videotaped with two separate cameras to ensure the best view possible of their faces.

The presence of infants' smiles and cry-faces in the FFSF were coded by coders certified in the Facial Action Coding System (FACS) (Ekman & Friesen, 1978) and trained in its application to infants, BabyFACS (Oster, 2000, Unpublished monograph and coding manual). Smiles and cry-faces are, respectively, infants' prototypical expressions of positive and negative emotion. The upward turning of the lip corners produced by the contraction of the zygomatic major defines smiling (AU12). In cry-faces, the lips are stretched laterally by the risorius muscle (AU20) and the brows are lowered by the corrugator muscle (AU4). Neutral affect consisted of the proportion of time in which the infant was not smiling or exhibiting a cry-face. Approximately 23% of infants were coded for reliability with mean 86% agreement between coders (mean $\kappa = .67$). The main and reliability coders were not blind to subject status.

FFSF episodes were terminated if the infant cried for >20 s or if the parent elected to terminate the episode. One parent of an ASD-sib elected to stop the FFSF procedure after the FF episode because her infant was upset. Thus, this child was included only in the FF portion of the analyses and not in the SF, RE, or elsewhere. The durations of the episodes can be found in Table 1. The FF episode was significantly shorter for ASD-sibs than TD-sibs $F(1, 29) = 4.41$, $p = .05$. There were no duration differences for the SF or RE episodes.

All analyses were conducted using the proportion of time spent either smiling, in a cry face, or with a neutral expression as the dependent variable. In order to calculate the proportion of time infants were engaged in neutral, smiling, and cry-face expressions during a given FFSF episode, the duration of the expression was divided by the duration of the episode excluding time during which the coder could not see the face of the child (see Table 2). We also created time-based (not episode-based) proportions of neutral, smiling, and cry-faces throughout the FFSF protocol (excluding time in which the coder could not see the face of the child). Doing so allowed us to include data from all infants enrolled in the study, including the child who completed only the FF portion of the FFSF. Because the FF and RE episodes had baseline durations of 3 minutes, and the still-face 2 minutes, the total proportion variables were not simply the sum of the proportions of smiling or cry-faces in each episode (see Table 2).

To examine stability of smiling and cry-faces across FFSF episodes, correlations were calculated for each group. These correlations were transformed to *Z*-scores to test for differences in the stability of inter-episode emotional expression between the two groups (Meng, Rosenthal, & Rubin, 1992).

All infants were administered the Early Social Communication Scales (ESCS) (Mundy et al., 1996) at least once between 8 ($M = 8.24$, $SD = .25$), 10 ($M = 10.41$, $SD = .28$), 12/13 ($M = 12.79$, $SD = .61$), 15 ($M = 15.41$, $SD = .43$), and 18 months ($M = 18.23$, $SD = .41$) of age. There were eight infants who participated in only one visit, four infants who participated in two visits, five infants who participated in five visits, seven infants who participated in four visits, and seven infants who participated in all five visits. The number of participants at each assessment can be found in Table 3. The ESCS is a semi-structured assessment which elicits early social communication, particularly joint attention and behavioral requesting, and the sharing of positive affect with an examiner. Parents are instructed not to initiate contact with their child while their child sits on their lap and is presented with attractive toys and objects by an examiner. The ESCS, which is designed for children between the ages of 8 and 24 months, takes approximately 15–25 minutes to administer.

Responding to joint attention refers to the child's ability to follow the joint attention behavior (i.e., pointing) of the examiner. RJA was coded when infants followed the examiner's point combined with a vocalization (i.e., the child's name) to a distal stimulus. IJA refers to a child's ability to share his or her interest or joy in an object. IJA was coded when infants made eye contact with the experimenter while manipulating a static or active toy or alternated eye contact between a distal, active mechanical toy and the experimenter. If an infant alternated eye contact after a mechanical toy became inactive, IJA episodes were only coded within 2 seconds of the time the toy became inactive. This ensured that the infant's behavior was indeed related to the object. Episodes in which the experimenter's overt behaviors (e.g., talking or moving) may have elicited the infant's attention were not coded.

Table 1 Duration (in minutes) of the face-to-face/still-face (FFSF) episodes

Episode	Autism Spectrum Disorders (ASD)-sibs			Typically developing siblings (TD-sibs)		
	n	Mean (*SD*)	Range	*n*	Mean (*SD*)	Range
Face to face (FF)	12	2.84 (.41)	1.90–3.04	19	3.04 (.06)*	2.96–3.18
Still face (SF)	11	1.98 (.16)	1.49–2.06	19	1.99 (.18)	1.38–2.42
Reunion (RE)	11	3.02 (.02)	2.97–3.04	19	2.68 (.73)	.50–3.07

* Significant group difference $p \le .05$

Table 2 Proportion of time spent smiling, in cry-faces, and in neutral affect in the face-to-face/still-face (FFSF)

Episode	Smile mean (*SD*)		*p* (d)	Cry-face mean (*SD*)		*p* (d)	Neutral affect mean (*SD*)		*p* (d)
	ASD	TD		ASD	TD		ASD	TD	
Face to face (FF)	.21 (.13)	.31 (.16)	.07 (.69)	.07 (.25)	.01 (.03)	.44 (–.34)	.71 (.21)	.67 (.16)	.51 (–.21)
Still face (SF)	.05 (.06)	.06 (.09)	.62 (.13)	.10 (.20)	.08 (.17)	.79 (–.10)	.85 (.20)	.85 (.17)	.95 (.00)
Reunion (RE)	.17 (.11)	.26 (.17)	.14 (.63)	.07 (.11)	.13 (.27)	.35 (.29)	.76 (.15)	.61 (.23)	.06 (–.77)
Total	.15 (.09)	.24 (.11)	.04 (.90)	.11 (.24)	.06 (.10)	.35 (–.27)	.79 (.11)	.71 (.14)	.12 (–.64)

Note: The total variable is not the sum of the proportions in each episode (which differ in length) but the proportion of either smiling, in cry-faces, or in neutral affect in the total time in the FFSF

Table 3 Mean levels of behaviors in the early social communication scales

Age	*n*		Total initiating joint attention (IJA)		*p* (d)	Lower initiating behavioral requests (IBR)		*p* (d)	Higher IBR		*p* (d)	Passed responding to joint attention (RJA) trials		*p* (d)
	ASD	TD	ASD	TD		ASD	TD		ASD	TD		ASD	TD	
8 Months	9	16	.80 (.44)	1.14 (.56)	.13 (.68)	.44 (.40)	.38 (.18)	.69 (–.19)	.07 (.06)	.12 (.15)	.28 (.44)	1.56 (1.59)	2.50 (1.46)	.15 (.62)
10 Months	8	14	1.35 (.55)	1.48 (.55)	.61 (.24)	.56 (.27)	.56 (.25)	.94 (.00)	.17 (.16)	.37 (.42)	.22 (.63)	2.38 (2.13)	1.93 (1.98)	.63 (–.22)
12/13 Months	6	10	1.58 (1.08)	1.15 (.49)	.30 (–.51)	.49 (.35)	.64 (.67)	.65 (.28)	.14 (.16)	.66 (.50)	.01 (1.40)	1.60 (1.14)	3.00 (2.79)	.19 (.66)
15 Months	5	12	.48 (.19)	.95 (.46)	.01 (1.34)	.43 (.49)	.48 (.25)	.77 (.13)	.32 (.32)	.77 (.67)	.18 (.86)	2.80 (1.79)	5.00 (2.66)	.11 (.97)
18 Months	6	10	1.13 (.59)	1.31 (.84)	.66 (.25)	.67 (.56)	.65 (.26)	.92 (–.05)	.51 (.35)	.83 (.32)	.10 (.95)	3.17 (2.14)	5.50 (1.78)	.03 (1.18)

There are both lower and upper level behaviors associated with IJA. Lower level IJA behaviors include gazing at an examiner while watching or holding an active toy, or making eye contact while holding or touching an inactive toy. Higher level IJA behaviors include pointing, with or without eye contact, at an object of interest or clearly holding up a toy and showing it to the examiner.

Episodes of IBR refer to requests for help or responses to a direct question. IBR occurs when the child requests something from another person or would like the person to do something for him or her. There are lower and higher level IBR behaviors. Lower level IBR behaviors include making eye contact to request a toy or reaching toward a toy, either with or without eye contact. Higher level IBR behaviors include pointing at a desired toy, either with or without eye contact, or giving the examiner a toy, either with or without eye contact.

The ESCS was coded by two main coders and one reliability coder. One of the main coders was blind to all but 2 infants' status, and the other main coder was blind to all but 13 infants' status. The reliability coder was not blind to subject status. Each main coder coded each child in at least one time point. Approximately 20% of infants were double coded by a reliability coder with mean intra-class correlations of ≥.70 for all behaviors coded including lower level IJA, higher level IJA, total IJA, lower level IBR, higher level IBR, total IBR, and RJA. The occurrence of each of these behaviors was expressed as a rate per minute with respect to the total duration of the ESCS with the exception of RJA which was expressed as the number of correctly followed trials (out of 8 trials). These variables were used in all subsequent analyses. Due to the low number of children present at every one of the longitudinal assessments (TD-sibs $n = 5$, ASD-sibs $n = 2$), we were unable to conduct a repeated measures ANOVA for the ESCS variables. Instead, we compared groups at each age using two-tailed tests with an alpha level of .05. Only approximately half of the sample was 2 years of age or older, the age of our first standardized developmental assessment, at time of writing. This precluded matching participants on developmental and cognitive measures (Shaked & Yirmiya, 2004).

Results

Face-to-Face/Still-Face

To screen for group and protocol differences in the FFSF, we used 2 (group) by 3 (episode) separate repeated measures Analysis of Variance (ANOVA). We conducted these analyses and their follow-ups using the proportion of time first in smiles, then in cry-faces, and finally in neutral as dependent measures. The use of these proportions corrects for differences in the duration of episodes between infants. The one child who did not complete the entire FFSF protocol was excluded from all repeated measures analyses but was included in the analyses focusing on the FF episode. The repeated measures ANOVA of smiles produced a still-face effect in that smiling decreased during the SF portion of the FFSF, $F(2, 28) = 5.01$, $p = .03$, $\eta^2_p = .14$ (see Fig. 1). There were no significant status or interaction effects, $ps > .40$. However, our planned time-based group contrast of the entire FFSF protocol indicated that ASD-sibs smiled significantly less than TD-sibs over the entire FFSF protocol $t(29) = 2.21$, $p = .04$, $\eta^2_p = .14$ (see Table 2). The planned follow-up analyses revealed a non-significant trend for ASD-sibs to smile less than TD-sibs in the FF portion of the FFSF only, $t(29) = 1.86$, $p = .07$, $d = .69$ (see Table 2).

A repeated measures ANOVA indicated that infants also showed a still-face effect such that time exhibiting cry-face expressions increased during the SF portion of the FFSF, $F(2, 28) = 4.96$, $p = .03$, $\eta^2_p = .16$, for all infants (see Table 2). There were no interaction or status effects for cry-face expressions.

In a 2 (group) by 3 (episode) design, there were no group differences in neutral affect (times in which the infant was not smiling or exhibiting a cry-face), $p = .12$(see Table 2 and Fig. 2). Our planned time-based group contrast of the entire FFSF protocol indicated that there was a trend for ASD-sibs to show more neutral affect than TD-sibs in the RE portion of the FFSF, but this effect was not significant, $t(28) = -2.00$, $p = .06$, $d = -.77$. There were no significant neutral affect differences between the groups over the course of the FFSF protocol.

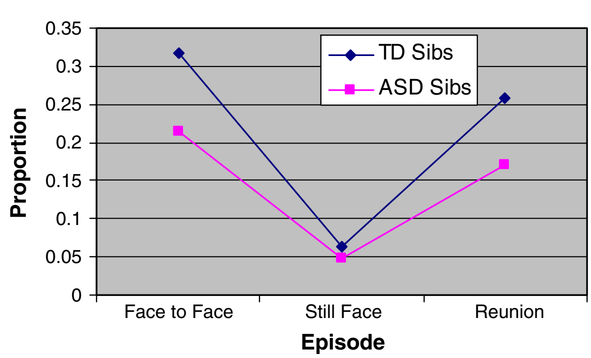

Fig. 1 Smiling in the face-to-face/still-face (FFSF)

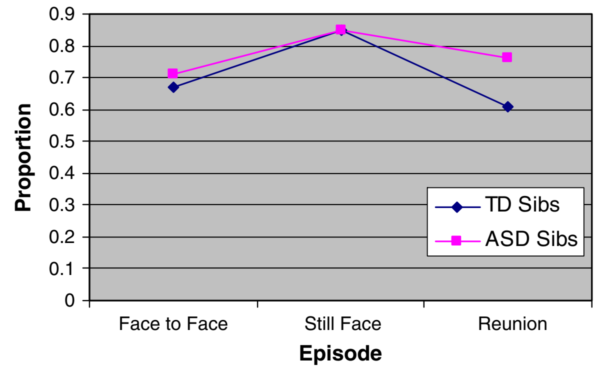

Fig. 2 Neutral affect in the face-to-face/still-face (FFSF)

Correlations of levels of expressive behaviors across the FFSF episodes revealed group differences (see Table 4). Both groups showed comparable levels of smiling stability between the FF and RE episodes. TD-sibs, but not ASD-sibs, showed stable levels of cry-faces between the FF and SF ($Z' = 2.49$, $p = .01$), and between the SF and RE episodes ($Z' = 2.00$, $p = .05$). ASD-sibs, but not TD-sibs, showed an unexpected positive correlation between cry-faces in the FF and smiling in the SF ($Z' = 2.46$, $p = .01$). These effects reflect both differences between the significance and non-significance of each group's correlations and significant differences between the correlation coefficients.

Early Social Communication Scales

The mean rate per minute of IJA in the ESCS can be found in Table 3. Preliminary analyses of higher-level and lower-level IJA revealed expected low rates of higher-level IJAs through 12 months. Results for lower-level IJAs mirrored those for total IJA, which are reported here. ASD-sibs had a smaller proportion of total IJA than TD-sibs at 15 months of age, $t(15.84) = 3.05$, $p < .01$, $d = 1.34$.[1] There were no status differences in IJA at other ages. There were also no group differences in rates of high and low IJA at any other age.

ASD-sibs responded to the examiner's joint attention bids (RJA) fewer times than TD-sibs at 18 months of age, $t(14) = 2.36$, $p = .03$, $d = 1.18$ (see Table 3). There were no status differences in RJA at other ages.

The mean rate per minute of IBR in the ESCS can be found in Table 3. ASD-sibs engaged in less higher level IBR at 12 months of age, $t(11.90) = 2.99$, $p = .01$, $d = 1.40$[1] There were no group differences for this variable at other ages.

[1] Levene's Test indicated a significant difference between group variances, prompting the correction evident in the non-integer *df*.

Table 4 Correlations between infant smiling and crying in the face-to-face/still-face (FFSF)

Episode	FF-smile	SF-smile	RE-smile	FF-cry	SF-cry	RE-cry
	Typically developing siblings (TD-sibs)					
	Autism Spectrum Disorders (ASD)-sibs					
Face to face (FF)-smile						
TD-sibs	_	.31	.54*	–.13	.07	–.05
ASD-sibs	_	–.19	.64*	–.51	.29	.01
Still face (SF)-smile						
TD-sibs	_	_	.31	*–.23*	–.25	–.24
ASD-sibs	_	_	–.04	.66*	–.12	–.39
Reunion (RE)-smile						
TD-sibs	_	_	_	–.40	–.31	–.53*
ASD-sibs	_	_	_	–.46	.32	–.09
FF-cry						
TD-sibs	_	_	_	_	*.70***	.36
ASD-sibs	_	_	_	_	–.17	–.25
SF-cry						
TD-sibs	_	_	_	_	_	*.76***
ASD-sibs	_	_	_	_	_	.16
RE-cry						
TD-sibs	_	_	_	_	_	_
ASD-sibs	_	_	_	_	_	_

* $p \leq .05$, ** $p \leq .01$

Note: An underlined correlation indicates that these effects reflect both differences between the significance and non-significance of each group's correlations and significant differences between the correlation coefficients at $p \leq .05$

Developmental Associations

In order to determine if there were associations among emotional expressions in the FFSF and indices of joint attention and related competencies in the ESCS, we correlated the total proportion of time spent smiling, exhibiting a cry-face, or in neutral affect in the entire FFSF protocol with average rates of IJA, RJA, and high and low IBR at all ages. There were no significant correlations for the ASD-sib group, the TD-sib group, or both groups combined.

Discussion

Infant siblings of children with autism are at increased risk for developing an ASD or deficits within the broad phenotype. Much research on the early deficits associated with ASD has been conducted retrospectively making it difficult to ascertain valid levels of early functioning. In this current prospective study, we examined the broad phenotype of ASD in the younger siblings (between the ages of six to eighteen months) of children diagnosed with ASD. We found evidence for some differences between ASD-sibs and TD-sibs as early as six months of age and inconsistent differences in IJA, RJA, and IBR.

At six months of age, ASD-sibs smiled for a significantly lower proportion of time during the FFSF protocol with their parent than did TD-sibs. There was a non-significant trend for ASD-sibs to exhibit more neutral affect than TD-sibs in the reunion portion of the FFSF. ASD-sibs engaged in significantly lower rates of IJA than TD-sibs in the ESCS at 15 months and in lower rates of higher-level IBR at 12 months. They also responded to the joint attention bids of the examiner (RJA) significantly less frequently than TD-sibs at 18-months of age.

Limitations of the study include its small sample size and the lack of ESCS measures from each participant at each age and lack of outcome data for the siblings. Not all infants in this sample have attained 18 months of age, and their longitudinal patterns of attendance for the ESCS administrations were variable. The small number of children present at each and every longitudinal assessment precluded the use of a repeated measures ANOVA for the ESCS variables. Multiple comparisons conducted for the FFSF variables and an alpha level of .05 increased the possibility that significant group differences might emerge by chance. We adopted these strategies so as to not overlook potentially important behavioral differences between ASD-sibs and TD-sibs in this initial sample. Nevertheless, results should be interpreted with caution and with respect to the growing body of research characterizing the developing ASD phenotype.

Using overall time in the FFSF as the metric for comparison, ASD-sibs smiled less than TD-sibs, a large effect size explaining 14% of the variance. An episode-based repeated-measures examination of these did not reveal this effect. This discrepancy reflects the increased impact of the FF and RE episodes of the protocol in the time-based analysis. It also reflects the influence of an ASD-sib who was excluded from the repeated-measures analysis because he cried but did not smile during the FF and did not complete the remainder of the FFSF protocol. Yirmiya et al. (2006) did not find smiling deficits in ASD-sibs at 4 months. The current results nevertheless mirror reports of positive affect attenuation in prospective and retrospective studies involving children diagnosed with an ASD (Baranek, 1999; Boelte & Poustka, 2003; Bolton et al., 1998; Capps et al., 1993; Murphy et al., 2000; Osterling et al., 2002; Wimpory et al., 2000; Yirmiya et al., 1989). Our findings are similar to parental reports of a reduction of emotionally positive behaviors during the play of children with ASD (Capps et al., 1993). Like Yirmiya et al. (2006), we found no differences in infants' mean levels of negative facial expressions during the FFSF.

There was a tendency for ASD-sibs to show greater neutral affect than TD-sibs during the reunion episode of the FFSF. This trend was non-significant and not independent of the smile and cry-face results (neutral affect was defined as their absence). Nevertheless, it is similar to Yirmiya et al.'s (2006) findings of greater ASD-sib affective neutrality during the still-face portion of the FFSF protocol. It is noteworthy that these findings of heightened neutral affect occurred in the typically stressful still face and reunion episodes, which present a challenge to the emotional regulation capacity of infants. Our findings are also consistent with observations of decreased smiling and of parent reports of passivity and a decreased activity level using the infant behavior questionnaire (IBQ) in 12-month-old ASD-sibs who proceeded to an ASD diagnosis (Zwaigenbaum et al., 2005). A growing literature suggests that reductions in affect expression manifested both as atypical neutrality and reductions in positive emotion are characteristic of the broad ASD phenotype in the first year of life.

TD-sibs showed continuity in facial expressions of negative affect between the FF and SF and between the SF and RE portions of the FFSF at four months. These continuities also have been documented in at-risk and normative samples (although gender differences with regard to emotional continuity have also been noted) (Acosta et al., 2004; Weinberg et al., 1999). ASD-sibs, however, had non-significant levels of negative emotional continuity and their levels of continuity were significantly lower than those of TD-sibs. Both groups showed continuity in levels of smiling between the FF and RE episodes. However, ASD-sibs who *smiled* more in the SF, a period of parental non-responsivity, showed greater *negative* affect when their parent re-engaged with them. These findings, combined with research suggesting that early ASD-related passivity is followed developmentally by increased distress and negativity (Zwaigenbaum et al., 2005), suggest a pattern of emotional instability and dysregulation. We did not find developmental continuities between emotional expression and referential communication difficulties, suggesting that much remains to be learned about the development of ASD-linked patterns of communicative development in the first 2 years of life.

Yirmiya et al. (2006) examined ASD-sib infant-mother synchrony in a free play FF episode, finding that synchrony was weaker in the SIBS-A dyads, only for infant-lead/mother-follow interactions. A limitation of the current study is that neither maternal expressions nor dyadic synchrony were examined. Nevertheless, it may be that ASD-sibs' potential emotional lability makes it more difficult for mothers to follow changes in their infants' moment-to-moment levels of emotional engagement and valence.

We investigated potential deficits in referential communication by examining IJA, RJA, and IBR during repeated administrations of the ESCS between 8 and 18 months of age. ASD-sibs initiated a lower rate of total IJA than TD-sibs at 15-months of age. Although this was a large difference accounting for 22% of the variance, no differences were found at other ages. Unreported analyses indicated a lack of significant differences in rates of high and low IJA behaviors at any age. Such inconsistent findings also characterize the existing literature. Goldberg et al. (2005) reported that ASD-sibs showed deficits in IJA and did not differ significantly from their older affected siblings. Yirmiya et al. (2006), however, found no IJA deficits at 14 months of age.

ASD-sibs responded to the joint attention bids of the examiner less than TD-sibs at 18-months of age, a large effect size accounting for 29% of the variance. Previous research has shown ASD-sibs in the second year of life have more difficulty locating the target of an adult's attention than do TD-sibs (Presmanes, Walden, Stone, & Yoder, 2007). Specific examinations of RJA in the ESCS in the first 2 years of life have yielded both reports of no differences (Yirmiya et al., 2006) and findings of an ASD-sib RJA decrement (Goldberg et al., 2005). The findings suggest that ASD-sibs may have difficulties in either understanding or responding to an adult's conventional request to orient attention to an object. These difficulties are consonant with deficits in children with ASDs who are less likely to respond to the examiner's joint attention bids than are typical children and children diagnosed with other developmental delays (Dawson et al., 2004).

Deficits in initiating and RJA bids are core deficits in children with ASD (Baranek, 1999; Dawson et al., 2004; Jones & Carr, 2004; Lewy & Dawson, 1992; Mundy et al., 1986; Werner et al., 2005). In early joint attention bids, infants share their interest in an object or event with another. In responding to these bids, infants manifest their understanding of conventional attempts to engage another's attention. The early—albeit inconsistent—differences in these abilities documented here may be important precursors to Theory of Mind (ToM) abilities. A decreased propensity to engage the attention of others and to respond to their bids for attention may contribute to deficits in the ability to understand mental states of others, a developing disability in individuals on the autism spectrum (Baron Cohen, Leslie, & Frith, 1985; Mundy, 1995).

ASD-sibs showed fewer higher-level behavioral requests than ASD-sibs at 12 months of age, a large effect accounting for more than 20% of the variance. Although we did not observe differences at 8, 10, 15, or 18 months of age, others have also observed deficits in requesting. Yirmiya et al. (2006) found that ASD-sibs engaged in fewer higher-level behavioral requests at 14-months of age than TD-sibs and Goldberg et al. (2005) found that ASD-sibs (14–19 months of age) engaged in less overall IBR than young TD children (10–19 months of age).

This paper contributes to a growing body of research suggesting deficits in requesting behaviors—particularly higher-level conventional requests such as points—among ASD-siblings during the second year of life. This may reflect difficulties in engaging socially to attain desired objects or acts. Specific ASD-sib deficits in generating conventional requests may index an inability to employ socially learned communicative behaviors, such as pointing, to achieve their goals. Such deficits are common among children on the autism spectrum (Osterling & Dawson, 1994) and may be an early manifestation of the tendency of children with ASD to use insufficiently referential and inappropriately physical requesting behaviors with others (Stone & Caro-Martinez, 1990).

In sum, ASD-sibs appear to show a range of inconsistent, but potentially related, difficulties with referential communication. These include subtle difficulties in IJA, later developing difficulties in responding to these joint attention bids, and deficits in requesting, particularly in conventional requests. These deficits in referential communication all occurred in the second year of life. A likely explanation is that such complex referential skills involving the more intentional use and comprehension of gaze and its coordination with more conventional gestures are crystallizing and developing during this period. Only as TD-sibs integrate these behaviors into their repertoires at stable levels in the second year of life are differences with ASD-sibs likely to emerge. Another possibility is that ASD-sibs may have a tendency toward passivity in interactions that is manifested in neutral affect in early infancy and scattered difficulties with IJA and IBR later in toddlerhood.

Heterogeneity among ASD-sibs is also likely to play a role in the inconsistent nature of the current findings. This preliminary study focused on group comparisons of infants with siblings on the autism spectrum and those with typically developing siblings. Unfortunately, only two of the ASD-sibs have completed the ADOS-G, a well-validated measure of ASD symptomatology (Lord, Rutter, DiLavore, & Risi, 1999). In addition, only approximately half of the sample is 2 years of age or older, the age of our first standardized developmental test (the Mullen Scales of Early Learning). As standardized measures of ASD severity are completed, we will investigate whether, within the ASD-sibs group, children who go on to an ASD diagnosis differ from children who do not attain this diagnosis but go on to show deficits associated with the broad ASD phenotype (i.e. language delay or sensory impairment) and whether such children, in turn, differ from ASD-sibs who develop none of these symptoms. It will be particularly important to determine if ASD-sibs who are later diagnosed with an ASD evidence differences in emotional expressions and rates of IJA, RJA, and IBR when compared to more typically developing ASD-sibs. Continued longitudinal study may also reveal ASD-sibs who show regression in development. Such infants may show typical development within the time frame considered in this study but show regressive increases in ASD symptomatology after 18 months of age. Some studies suggest that 20–40% of cases follow this developmental trajectory (Kobayashi & Murata, 1998; Maestro et al., 2002; Rogers, 2004).

We have noted subtle deficits in early emotional expression and later referential communication abilities in ASD-sibs that may index important early difficulties in emotional communication associated with the broad phenotype of ASD. We did not find developmental continuities between emotional expression and referential communication difficulties, suggesting that much remains to be learned about the development of ASD-linked patterns of communicative development in the first 2 years of life. Recent research suggests that children diagnosed with an ASD who undergo *early* and *intensive* intervention have better outcomes when compared to individuals who receive treatment later in life (Kazdin & Weisz, 2003). This paper contributes to a growing body of literature suggesting subtle, communicative deficits for ASD-sibs in the first 2 years. Determining the links between the characteristics of the broad phenotype in the first 18 months of life and diagnosable ASD that can benefit from intervention remains a topic for continued research.

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