Published in final edited form as:
Autism. 2008 September ; 12(5): 473–485. doi:10.1177/1362361308094504.

Visual disengagement in the infant siblings of children with an Autism Spectrum Disorder (ASD)

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Abstract

Children with Autism Spectrum Disorders (ASDs) are impaired in visually disengaging attention in both social and non-social contexts, impairments that may, in subtler form, also affect the infant siblings of children with an ASD (ASD-sibs). We investigated patterns of visual attention (gazing) in six-month-old ASD-sibs (n = 17) and the siblings of typically developing children (COMP-sibs; n =17) during the Face-to-Face/Still-Face Protocol (FFSF), in which parents are sequentially responsive, nonresponsive, and responsive to their infants. Throughout the protocol, ASD-sibs shifted their gaze to and from their parents' faces less frequently than did COMP-sibs. The mean durations of ASD-sibs' gazes away from their parents' faces were longer than those of COMP-sibs. ASD-sibs and COMP-sibs did not differ in the mean durations of gazes at their parents' faces. In sum, ASD-sibs showed no deficits in visual interest to their parents' faces, but greater interest than COMP-sibs in non-face stimuli.

Keywords

autism spectrum disorders; siblings; at-risk; disengagement; early deficits

Introduction

In early childhood, the ability to engage and disengage attention is necessary to the development of social communication. Engaging attention involves sustaining visual attention on an object, while disengaging attention involves shifting visual attention from one object to re-engage on another. Children with Autism Spectrum Disorders (ASDs) are able to sustain attention, but appear overly focused and impaired in their ability to disengage and shift attention (Landry & Bryson, 2004; Lovaas & Schreibman, 1971; Rincover & Ducharme, 1987; Wainwright Sharp & Bryson, 1993; Wainwright & Bryson, 1996). The purpose of this paper is to identify possible disengagement deficits associated with the broader autism phenotype and potential later ASD classification within the first year of life by examining the at-risk infant siblings of children with an ASD (ASD-sibs).

ASDs are pervasive developmental disorders characterized by impairments in social functioning, communication, and the display of repetitive behaviors and/or stereotyped interests (APA, 2000). Some of these associated deficits are also present, in the form of a broader phenotype, in the relatives of those with an ASD (Constantino et al., 2006; Dawson

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et al., 2004). The broader phenotype refers to milder deficits in at least one of the three affected domains in autism (Dawson et al., 2002). While 3 to 9% of full-siblings exhibit deficits associated with an ASD diagnosis, as many as 20% of full-siblings may exhibit deficits in social and communicative functioning, indicative of the broader phenotype (Landa & Garrett Mayer, 2006; Gamliel, Yirmiya, & Sigman, 2007).

Although ASDs are rarely diagnosed before two years of age, 30–50% of parents report prior abnormalities in their child's behavior from before the time of diagnosis (Werner, Dawson, Osterling, & Dinno, 2000; Zwaigenbaum et al., 2005). Early orienting and gaze-related deficits in autism have been identified through retrospective studies using parent report and analysis of early home videos. In home videos of ASD and typically developing (TD) children at eight and ten months of age, Werner et al. (2000) found that children with an ASD oriented to their name half as often as TD children, and were less likely than TD children to coordinate smiling with gaze at another person's face. Other retrospective studies using parent report have found similar deficits in orienting (Gomez & Baird, 2005; Osterling & Dawson, 1994; Zakian, Malvy, Desombre, Roux, & Lenoir, 2000). Overall, children with ASDs often display impairments that may be associated with disengagement deficits within the first year of life. These deficits may underlie difficulties in coordinating communication behaviors.

Landry and Bryson (2004) compared the visual-spatial attention (e.g., ability to disengage) of children with an ASD, children with Down syndrome, and TD children using a visual orienting paradigm. Children with an ASD disengaged significantly slower and had significantly fewer "fast" (< 300 msec) reaction times than the other two groups. Similarly, Newell et al. (2007) found that children with ASDs were impaired in shifting their attention when concurrent stimuli were present. These results indicate that children with ASDs have "sticky" visual attention which causes them to disengage more slowly or not disengage at all. This impaired disengagement may explain ASD individuals' deficits in eye contact, orienting to name, and joint attention in early development.

Recently, prospective studies have compared ASD-sibs with the infant siblings of typically developing children (COMP-sibs) (Yirmiya et al., 2006; Zwaigenbaum et al., 2005; Cassel et al., 2007) as ASD-sibs are at greater risk for developing an ASD or showing deficits associated with the broader autism phenotype (Landa & Garrett Mayer, 2006; Zwaigenbaum et al., 2005). Some of these studies have investigated possible early deficits in disengaging visual attention. Yirmiya et al. (2006) examined gaze patterns in four-month-old ASD-sibs and COMP-sibs during the Face-to-Face/Still-Face Protocol (FFSF) with the parent. ASD-sibs did not significantly differ from COMP-sibs in the proportion of time spent gazing at their mothers' faces. Additionally, Merin, Young, Ozonoff and Rogers (2007) found that six-month-old ASD-sibs did not differ from controls in the proportion of time they spent looking at their parents' faces versus away during the FFSF. However, Zwaigenbaum et al. (2005) found that those ASD-sibs whose disengagement difficulties increased from 6 to 12 months of age went on to receive a provisional ASD diagnosis at 24 months.

The current study further investigated disengagement by comparing six-month-old ASD-sibs' and COMP-sibs' gaze patterns at their parents' faces during the FFSF. Our goal was to examine infants' durations of engagement and disengagement at the parents' faces, and not simply overall time gazing at the parents' faces. We assessed visual attention through the FFSF, which has two periods of naturalistic interaction separated by a period of parental nonresponsivity. We hypothesized that ASD-sibs would shift their gaze to and from their parents' faces less frequently than COMP-sibs, and that they would have greater mean durations of gaze at and away from their parents' faces than COMP-sibs. Since the interactive episodes of the FFSF protocol are more social than experimental visual attention paradigms, we also examined the potential impact of parent behaviors, such as tickling, touching, and positive affect on infants' gaze patterns.

Method

Participants

All infants were approximately six-months old (*M* = 6.12, *SD* =.37) when they participated in the FFSF. Infants in this study were part of a larger sample investigating the social, emotional, and cognitive development of ASD-sibs and COMP-sibs. The present study included a sample of 34 infant-parent dyads (32 infant-mother dyads, 2 infant-father dyads); two of the ASD female infants were monozygotic twins. See Table 1 for the gender breakdown of ASD-sibs and COMP-sibs. Infants were included in the ASD-sibs group if the parent reported that at least one older sibling was diagnosed with Autism, Asperger's Syndrome, or Pervasive Developmental Disorder – Not Otherwise Specified (PDD-NOS), and diagnosis was confirmed through ADOS-G, Autism Diagnostic Interview-Revised (ADI-R; Lord, Rutter, & Couteur, 1994), and clinical impression. Infants were included in the COMP-sibs group if they had older sibling(s) not diagnosed with, and not showing any research evidence of, an ASD-related disorder (i.e. did not exceed cut off score of nine on the Social Communication Questionnaire (SCQ; Berument, Rutter, Lord, Pickles, & Bailey, 1999). Groups were comparable on gestational age (weeks), ethnicity, and parent education (see Table 1).

Face-to-Face/Still-Face Protocol

All six-month-olds participated in the FFSF Protocol (Tronick et al., 1978; Adamson & Frick, 2003), which consists of three episodes: the Face-to-Face (FF) episode, the Still-Face (SF) episode, and the Reunion (RE) episode. Parents were instructed to play with their infant (without toys) for three minutes (FF), then stop playing and hold a still-face for two minutes (SF), and resume play for another three minutes (RE). A two-second tone sounded at the beginning of each episode to notify the parents when a new episode had begun. The dyadic interaction was videotaped with a camera directed at the infant's face, a camera directed at the parent's face, and a camera that captured both infant and parent interacting. Infants were placed in a car seat and the parent was seated in a chair directly in front of the infant. FFSF episodes were terminated if the infant cried for more than 20 seconds or if the parent elected to terminate the episode. There were no group differences in the mean duration (minutes) of the FF (*M* = 3.00), SF (*M* = 1.97), and RE (*M* = 2.78). Twenty eight of the infant-parent dyads presented in the prospective study by Cassel et al. (2007) are included in the current study's sample. They found that during FFSF, six-month-old ASD-sibs smiled for a lower proportion of time than COMP-sibs.

Coding Gaze

Infants were coded as either gazing directly at the parent's face or not gazing at the parent's face. Thirty-eight percent of the video clips were randomly selected and coded by two coders (mean agreement = 90%; *kappa* = .75). The coders were blind to participant status.

Gaze patterns were examined using the overall frequency of gaze shifts and mean durations of gaze. The Frequency of Gaze Shifts was calculated as the sum of the frequency of gaze shifts to and away from the parent's face per minute; we did not separately examine gaze shifts at the parents face and away from the parents face because these values are, by definition, close to identical. The Mean Duration of Gaze At Parent was calculated as the mean duration of individual gazes at the parent's face. The Mean Duration of Gaze Away was calculated as the mean duration of individual gazes away from the parent's face. In exploratory analyses, we also calculated the Total Proportion of Gaze At Parent as the proportion of the number of frames in which the infant gazed directly at the parent's face out of the total number of frames. Data were screened for outliers (values two or more standard deviations above the mean). In supplementary analyses, outliers (never more than two per episode) were replaced with a data value one percent larger than the next most extreme non-outlier score in the distribution (Tabachnick & Fidell, 1996). A logarithmic transformation was performed to normalize the raw frequency of gaze shift and mean duration. The raw variables are presented in the figures and the logarithmically transformed variables were used in the statistical analyses. Neither the outlier procedure nor the log transforms impacted the significance levels reported below.

Coding Parent Behaviors

Due to the interactive aspects of the FFSF, parent behaviors that could potentially influence infants' gaze, such as tickling, touching, and smiling were also coded. Parent smiling at their infant during the FFSF was coded by a coder certified in Facial Action Coding System (FACS) (Ekman & Friesen, 1978). The onset of tickling occurred when the parent began moving their fingers while touching the infant. The offset of tickling occurred when the parent stopped moving their fingers and removed their hands from the child. Touch was coded when the parent made physical contact that did not involve tickling with the infant. Approximately 12% of the video clips were randomly selected and coded by two coders with a mean agreement of 88% (mean *kappa* = .81). Independent t-test showed that there were no differences between the groups on any of the three parent behaviors, $ps \geq .40$. The three types of parent behaviors were included as covariates in all repeated-measures ANOVAs.

Results

We compared the Frequency of Gaze Shifts (rate per minute) and the Mean Duration of Gaze Away and Gaze At Parent (seconds) of ASD-sibs ($n = 17$) and COMP-sibs ($n = 17$) across the FFSF.

A 2 (group) × 3 (episode) × 2 (gender) repeated-measures ANOVA, controlling for parent behaviors, indicated that ASD-sibs ($M = 13.19$ shifts; $SD = 5.55$) shifted their gaze to and from their parents' faces less frequently than COMP-sibs ($M = 18.16$ shifts; $SD = 8.36$), $F(1, 27) = 4.98$, $p = .03$, $\eta^2 = .16$ (see Figure 1). A non-significant interaction between group and gender indexed a tendency for female ASD-sibs to shift their gaze less frequently than female COMP-sibs, and male ASD- and COMP-sibs $F(1, 27) = 3.11$, $p = .08$, η2 = .11. Within-subjects contrasts indicated that infants, regardless of group, shifted their gaze at their parents' faces less frequently during the SF compared to the FF and RE, $F(1, 27) = 4.28$, $p = .05$, $\eta^2 = .14$. There was no interaction between episode and group, $F(2, 27) = .84$, $p = .41$. There were no gender main effects.

To follow-up on the group differences in the Frequency of Gaze Shifts, we examined group differences in Mean Duration of Gaze At Parent and Mean Duration of Gaze Away. First, we examined the association of these variables for the entire sample. The Mean Duration of Gaze Away was not correlated with Mean Duration of Gaze At Parent in any of the episodes in the FFSF (FF $r = -.20$; SF $r = -.21$; RE $r = -.18$). The Frequency of Gaze Shifts was negatively correlated with Mean Duration of Gaze Away in each episode of the FFSF (FF $r = -.57$; SF $r = -.93$; RE $r = -.62$), $ps < .01$, and with the Mean Duration of Gaze At Parent during the FF ($r = -.62$) and RE ($r = -.61$), $ps < .01$, but not the SF ($r = -.11$). Thus there was not a one-to-one correspondence between these variables, although a higher Frequency of Gaze Shifts was associated with briefer durations of individual gazes.

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A 2 (group) × 3 (episode) × 2 (gender) repeated-measures ANOVA of Mean Duration of Gaze Away, controlling for parent behaviors, indicated that ASD-sibs (*M* = 8.44 seconds; *SD* = 16.15) had significantly longer Mean Duration of Gaze Away than COMP-sibs (*M* = 4.72 seconds; *SD* = 4.00) across the FFSF protocol, $F(1, 27) = 5.31$, $p = .03$, $\eta 2 = .16$ (see Figure 2). A non-significant interaction between group and gender indexed a tendency for female ASD-sibs to have longer Mean Duration of Gaze Away than female COMP-sibs, and male ASD- and COMP-sibs, $F(1, 27) = 3.23$, $p = .08$, $\eta 2 = .11$. There was no interaction between episode and group, $F(2, 27) = .56$, $p = .58$. There were no significant gender or episode main effects.

A 2 (group) × 3 (episode) × 2 (gender) repeated-measures ANOVA of Mean Duration of Gaze At Parent, controlling for parent behaviors, indicated that there were no differences between ASD-sibs (*M* = 4.16 seconds; *SD* = 3.33) and COMP-sibs (*M* = 3.47 seconds; *SD* = 2.35) in Mean Duration of Gaze At Parent across the FFSF protocol $F(1, 27) = .03$, $p = .95$, $\eta 2 = .00$. There was no interaction between episode and group, $F(2, 27) = 1.20$, $p = .31$. There were no significant gender, episode, or interaction effects.

The Total Proportion of Gaze At Parent was, unsurprisingly, strongly positively correlated with Mean Duration of Gaze At in each episode of the FFSF (FF $r = .74$; SF $r = .65$; RE $r = .66$), $ps < .01$, and strongly negatively correlated with Mean Duration of Gaze Away (FF $r = -.75$; SF $r = -.86$; RE $r = -.80$), $ps < .01$. To increase comparability with previous reports, we, nevertheless, conducted a 2 (group) × 3 (episode) × 2 (gender) repeated-measures ANOVA of Total Proportion of Gaze At Parent, controlling for parent behaviors. There were no group differences in Total Proportion of Gaze At across the FFSF protocol, $F(1, 27) = 2.67$, $p = .12$, $\eta 2 = .09$.

Discussion

Over 60% of parents report, retrospectively, that their autistic children, “looked through or past people” in the first year of life (Gomez & Baird, 2005, pg. 113). The current study is one of several attempting to prospectively identify potential early deficits associated with the broader autism phenotype as well as the later diagnosis of an ASD in the infant siblings of children with ASDs (Yirmiya et al., 2006; Zwaigenbaum et al., 2005; Landa & Garrett Mayer, 2006). We found that, during the FFSF, ASD-sibs shifted their gaze less frequently and had longer mean durations of gaze away from their parents’ faces than COMP-sibs.

Landry and Bryson (2004) found that older children with Autism/PDD took significantly longer to disengage from experimental stimuli than both TD children and children with Down Syndrome. Using a visual-orienting paradigm, Zwaigenbaum et al. (2005) found that ASD-sibs, who became slower at disengaging from standard stimuli between six and twelve months of age, went on to receive an ASD classification on the ADOS at 24 months of age. There may be a neurological basis for these disengagement deficits given that delayed attentional orienting has been associated with severity of cerebellar hypoplasia in autistic children between two and six years of age (Harris et al., 1999). ASD-sibs had a lower frequency of gaze shifts to and from their parents’ faces and longer mean duration of gazes away from their parents’ faces than COMP-sibs, which may represent developing deficits in the capacity to disengage, which characterize the hallmark “sticky” attention of children with ASDs.

Merin et al. (2007) used the FFSF procedure to assess infant-parent visual and emotional engagement during a dyadic interaction and perturbations of that interaction. They did not find differences in the proportion of time six-month-old ASD-sibs and COMP-sibs gazed at versus away from their parents’ faces. Yirmiya et al. (2006) also found no differences between six-month-old ASD-sibs and COMP-sibs in the proportion of time they gazed at their parents' faces throughout the FFSF. We replicated these null effects for proportion of time gazing at the parent. Relatedly, we found no differences between the groups in Mean Durations of Gaze At Parent. Effect sizes for these nonsignificant analyses were near zero, suggesting, in fact, that ASD-sibs do not show reduced gazing at their parents.

Compared to COMP-sibs, ASD-sibs had longer mean durations of gazing away from their parents' faces. This suggests that while ASD-sibs are more interested in non-social stimuli than COMP-sibs, both sibling groups are equally interested and engaged by the parent's face, a salient social stimulus. The findings may be relevant to children with ASDs. These children are delayed in their ability to shift attention and engage with social stimuli (Dawson, Meltzoff, Osterling, Rinaldi, & Brown, 1998; Dawson et al., 2004), but, once engaged with social stimuli like their parents' faces, may do so for a typical amount of time. Sigman, Mundy, Sherman, and Ungerer (1986), for example, found that older children with an ASD spent a similar amount of time gazing at their caregivers when compared to TD and children with mental retardation.

This study expands our understanding of potential deficits that may be associated with the broad phenotype or increase vulnerability to developing ASDs. Atypical patterns of disengagement may have adverse implications for emotional regulation, orienting, joint attention, and other abilities directly associated with socio-emotional functioning. The ability to disengage visual attention is important in the regulation of emotional states (Rothbart, Ziaie, & O'Boyle, 1992). Children at risk for autism may exhibit a tendency to fussiness (Yirmiya et al., 2006) due to their inability to easily disengage from a disconcerting interaction (Bryson et al., 2004).

Deficits in shifting gaze and orienting may also be related or lead to deficits in joint attention abilities, as the former may be a rudimentary, but necessary mechanism for the emergence of the latter (Mundy et al., 2007, Morales et al., 2000). Initiating joint attention (IJA) requires using gaze to communicate about an object or event in the environment (Jones & Carr, 2004; Messinger & Fogel, 1998). Respond to joint attention requires following the joint attention (e.g., pointing) of another individual. In both circumstances, children must disengage from one object or event to either initiate joint attention or respond to joint attention bids. Deficits in IJA and RJA have been found in children with autism (Baranek, 1999; Jones & Carr, 2004) and in ASD-sibs (Goldberg et al., 2005; Cassel et al., 2007), and have helped differentiate the majority of children with an ASD from children with other developmental delays (Lewy & Dawson, 1992; Mundy, Sigman, Ungerer, & Sherman, 1986). Emerging impairments in visual attention such as those documented here may predict later deficits in joint attention in children at-risk for an ASD. We did not, however, contrast participants based on later ASD classification because 15 out of the 34 participants had not reached 30 months of age.

Overall, ASD-sibs' decreased ability to shift their attention to social presses may be due to their heightened interest in less social, non-face stimuli in their environment. These findings extend our knowledge of visual attention differences among infants at risk for autism in the first year of life. The differences may be representative of early deficits and could have adverse implications for developmentally critical domains such as emotional regulation, joint attention, and other abilities directly associated with socio-emotional functioning.

Acknowledgments

We wish to thank the families for participating in this research. Funding for this study was provided by grants from NICHD (047417), Autism Speaks, and the Marino Autism Research Institute.

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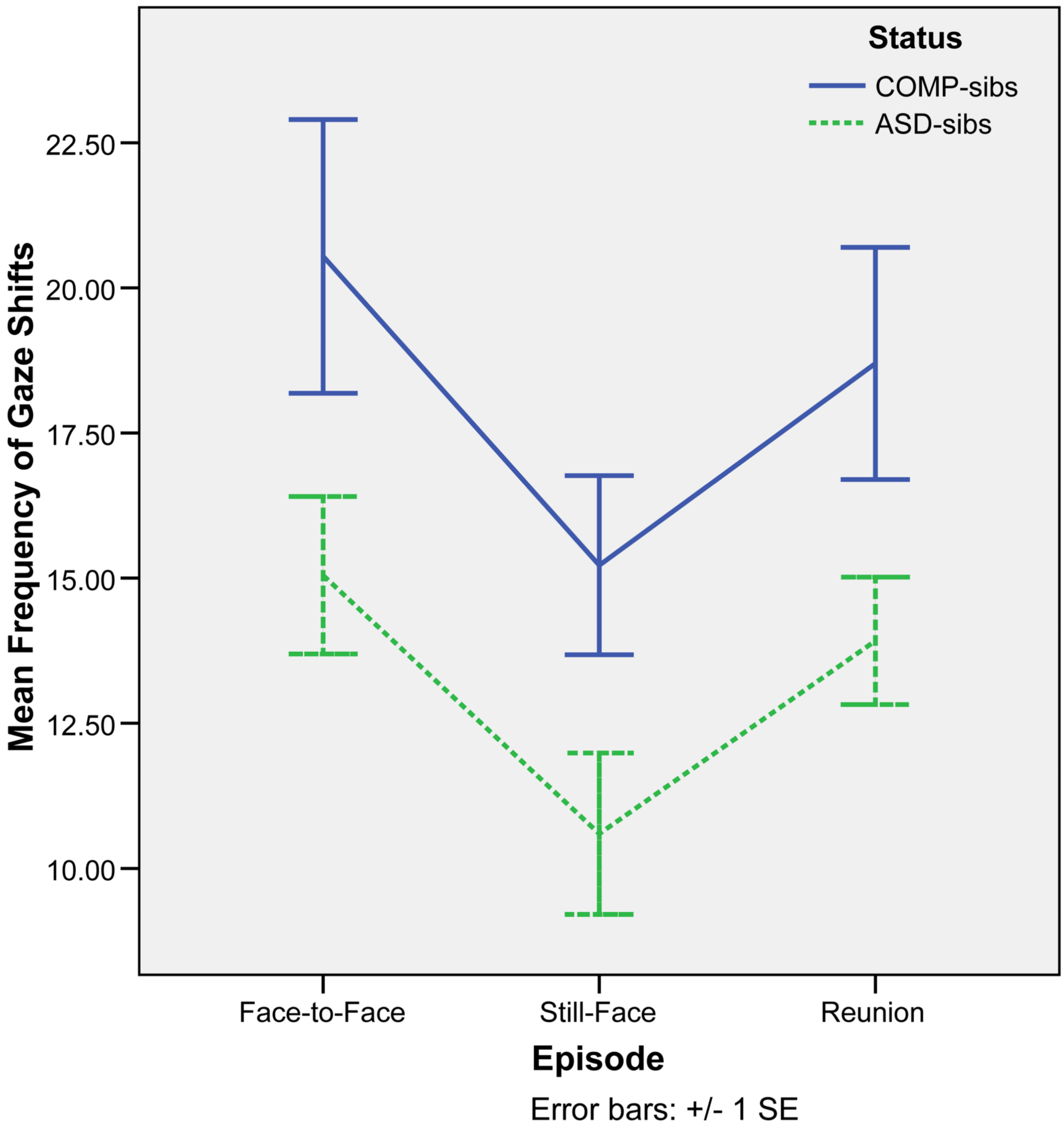

Figure 1. Overall Gaze Shifts in the FFSF

Note: ASD-sibs has significantly lower Frequency of Gaze Shifts than COMP-sibs across the FFSF protocol, $F(1, 27) = 4.98$, $p = .03$, $\eta 2 = .16$.

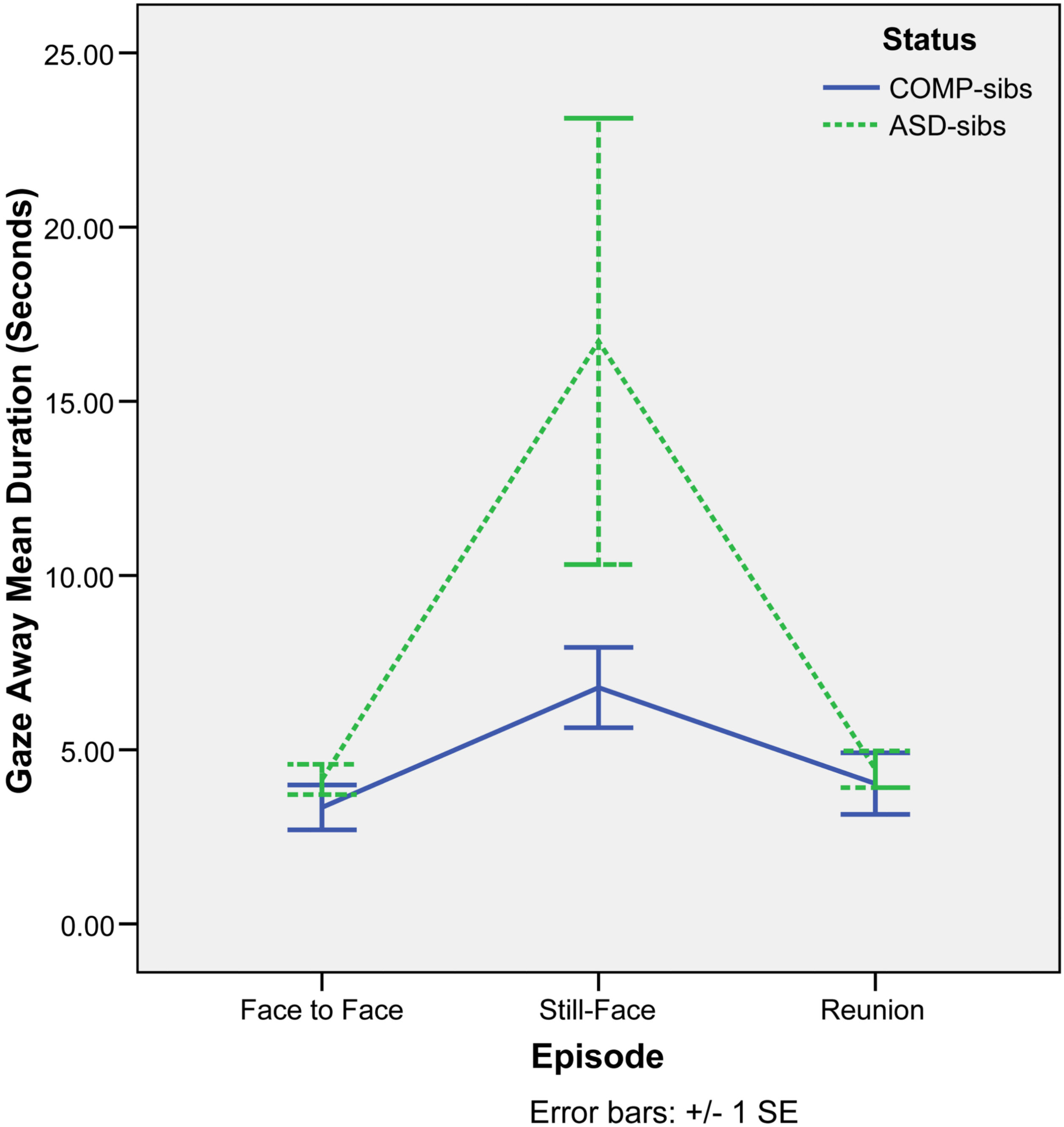

Figure 2. Gaze away from parent's face (mean duration) during the FFSF

Note: ASD-sibs had significantly longer Mean Duration of Gaze Away than COMP-sibs across the FFSF protocol, $F(1, 27) = 5.31$, $p = .03$, $\eta 2 = .16$. There was no interaction between episode and group, $F(2, 27) = .56$, $p = .58$.

Table 1

Participant Demographics

Demographics	ASD-sibs	COMP-sibs
Gender [% / (n)]		
Male	58.8 / (10)	47.1 / (8)
Female	41.2 / (7)	52.9 / (9)
Ethnicity [% / (n)]		
White/Non-Hispanic	29.41 / (5)	35.29 / (6)
White/Hispanic	70.59 / (12)	41.17 / (7)
African-American/Biracial	0 / (0)	17.64 / (3)
Asian	0 / (0)	5.88 / (1)
Parent Education [% / (n)]		
Some College	11.6 / (2)	29.4 / (5)
4-year College	23.5 / (4)	29.4 / (5)
Advanced Professional Degree	64.7 / (11)	41.2/ (7)
Gestational Age (Weeks) [M / (SD)]	39.06 / 1.20	38.47 / 1.46

Note: There were no significant differences between ASD-sibs and COMP-sibs on these demographics variables.