JSLHR

Research Article

Joint Engagement and Early Language in Young Children With Fragile X Syndrome

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Purpose: In this study, we examine joint engagement (JE) in young children with fragile X syndrome (FXS) and its relationship to language abilities and autism spectrum disorder symptomatology at 24 to 36 months (toddler period) and 59 to 68 months (child period).
Method: Participants were 28 children with FXS (24 boys, four girls) and their mothers. Videotaped home observations were conducted during the toddler period and coded for JE. Language abilities were measured at both ages from a developmental assessment, a functional measure, and from a language sample. The Childhood Autism Rating Scale (Schopler, Reichler, & Renner, 1988) was completed at both ages.
Results: Children with FXS spent more time in supported JE than in coordinated JE. Using a weighted JE variable, we found that children with FXS who had higher weighted JE scores also had more advanced expressive language skills at both the toddler and child periods. Weighted JE was negatively related to autism symptomatology in the toddler period.
Conclusion: This study provides evidence that children with FXS who use more JE also have more advanced expressive language skills in early development. Therefore, existing early interventions that target JE behaviors may be effective for promoting language, social communication, and social interaction in this population.

Early social behavior serves as an especially important foundation for language acquisition in children with intellectual and developmental disability (IDD; Abbeduto, Evans, & Dolan, 2001). Examining early social behaviors that underlie language development is important for children with fragile X syndrome (FXS) because they typically have impairment in both language and social development (Abbeduto, Brady, & Kover, 2007). Hence, examining these early behaviors may provide insight into the cascading effects of these social experiences and behaviors on subsequent language development of these children (Fidler, Lunkenheimer, & Hahn, 2011) and identify potential targets for early intervention. Despite their importance, however, early social behaviors have not been closely examined in young children with FXS. Thus, the purpose of the present study is to examine joint engagement (JE)—an early social behavior—in young children with FXS.

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Editor: Rhea Paul
Associate Editor: Linda Watson

Received January 5, 2015
Revision received May 26, 2015
Accepted February 9, 2016
DOI: 10.1044/2016_JSLHR-L-15-0005

FXS is the most common inherited cause of developmental disability (Crawford, Acuna, & Sherman, 2001). FXS results from a mutation of the *fragile X mental retardation-1* gene on the X chromosome (Verkerk et al., 1991). Because of this, boys have a higher incidence rate than girls—one in 4,000 boys versus one in 6,000 girls—and are typically more severely affected (Centers for Disease Control and Prevention, 2011; Hagerman, 2007). There is a wide range of effects for individuals with FXS, from learning disabilities to severe intellectual disabilities (Loesch, Huggins, & Hagerman, 2004). FXS is also the most common genetic cause of autism spectrum disorder (ASD; Hagerman, Rivera, & Hagerman, 2008) with 30%–50% of individuals meeting diagnostic criteria for autism and 60%–74% meeting criteria for ASD (this includes those meeting criteria for pervasive developmental delay; Bailey, Raspa, Olmsted, & Holiday, 2008; Clifford et al., 2007; Hall, Lightbody, Hirt, Rezvani, & Reiss, 2010; Hall, Lightbody, & Reiss, 2008; Harris et al., 2008; Kaufmann et al., 2004). There is a unique behavioral phenotype associated with FXS indicating a distinct pattern of relative strengths and weakness (for a review, see Hagerman, 2007). However, most of this research has focused on older children, adolescents, and adults. Examining early development, especially early social development, has important implications for language development and early identification of comorbid ASD in this population.

Disclosure: The authors have declared that no competing interests existed at the time of publication.

Early Social Development in Infants and Young Children

Over the course of the first year of life, infants begin to engage with the world. First, infants engage in face-to-face interactions with their caregivers, then with objects, and last, they begin to interact with caregivers around objects of interest (Trevarthen & Aitken, 2001). These behaviors continue to build in complexity and utility over the course of early childhood. There is general consensus that early social experiences, including parent–child interactions, form a critical foundation for language, cognitive, and social development (Adamson & Bakeman, 1991; Trevarthen & Aitken, 2001). One way to characterize early social behavior is to examine the child's engagement with people, events, and objects within the context of a naturalistic social interaction with their mothers.

Engagement States

An *engagement state* refers to a child's active attention to objects or people (Adamson & Bakeman, 1991; Bakeman & Adamson, 1984), which is typically measured in terms of a distinct period of time (usually seconds or minutes). Engagement states, then, describe episodes of a child's engagement or lack of engagement with a person, an object, or both an object and another person but not a specific set of behaviors, such as eye gaze or gestures to social partners (see Table 1 for examples; Adamson & Bakeman, 1991).

Instances of JE with a caregiver around an object can be divided into two forms: *supported joint engagement* (SJE) and *coordinated joint engagement* (CJE). In SJE, the child and caregiver are engaged with the same object, but the child is not actively and reciprocally responding to the caregiver (Adamson, Bakeman, & Deckner, 2004; Bakeman & Adamson, 1984). The child's emerging behaviors of sharing interest about an object lead the caregiver to scaffold the child's engagement with the object and, therefore, take on the role of maintaining an interaction around a shared interest (Adamson & Bakeman, 1991; Bakeman & Adamson, 1984). In contrast, during periods of CJE, the child actively engages with either an object or event of interest and the caregiver (Adamson et al., 2004; Bakeman & Adamson, 1984). In this form of JE, there is a dynamic and reciprocal alternating of attention and sharing of interest between the object and the caregiver (Adamson et al., 2004; Bakeman & Adamson, 1984; see Table 1 for examples).

Developmental Progression of Engagement States

In the first month of life, typically developing infants will visually attend, briefly, to people, objects, and events within their social context (Adamson & Chance, 1998; Trevarthen & Aitken, 2001). During the second month, infants begin to focus their visual attention on primarily engaging in face-to-face interactions with a social partner, usually their caregiver, with the purpose of sharing emotional expressions with one another (i.e., person engagement state; Adamson & Chance, 1998; Trevarthen & Aitken, 2001). Between 5 and 6 months, infants shift their attention to objects in their environment (i.e., object engagement state). Although this may decrease their attention to the caregiver, periods of person engagement will continue to occur (Adamson & Chance, 1998; Trevarthen & Hubley, 1978). In addition, during these instances of object engagement, the caregiver may join in the infant's engagement with an object, leading to periods of SJE (Adamson & Bakeman, 1991; Adamson & Chance, 1998).

The first instances of both SJE and CJE can be observed in typically developing infants at 6 months (Adamson & Chance, 1998; Bakeman & Adamson, 1984). At this time, most interactions with a caregiver involve prolonged periods of SJE that are punctuated by brief moments in which the infant will look between the object and the caregiver, marking the first instances of CJE (Adamson & Chance, 1998, Bakeman & Adamson, 1984). The caregiver's scaffolding at this time helps to maintain the periods of SJE. In the following months, infants will consolidate the behaviors of shifting attention between an object and a caregiver, leading to sustained periods of CJE at about 13 months (Adamson & Chance, 1998; Bakeman & Adamson, 1984). Infants continue to engage in SJE and CJE and by the middle of the second year typically execute CJE with ease when interacting with a caregiver (Adamson & Chance, 1998).

Table 1. Definition of engagement state codes.

Engagement state	Definition	Example
Unengaged	The child is uninvolved with people or objects.	The child is sitting on the floor scanning the room.
Person	The child is engaged with the mother in a face-to-face interaction.	The child and mother are playing peek-a-boo.
Object	The child is engaged with an object and only attending to the object.	The child is focused on stacking a set of blocks.
Supported joint engagement	The child and the mother are engaged with the same object, but the child is not actively responding to the mother's participation.	The mother and child are playing with a puzzle together, and she encourages the child's play by saying, "Where does that piece go?" The child puts the piece in place, but does not look at or respond to the mother.
Coordinated joint engagement	The child and the mother are engaged with the same object, and the child is actively and repeatedly acknowledging the mother's participation with the object.	The mother and child are playing with a Jack-in-the-box, and when Jack pops out, the child looks back and forth between the mother and the toy.

Although there is a developmental sequence for engagement states, infants and young children will continue to use all forms of engagement (i.e., person, object, SJE, and CJE) once they have integrated those skills, depending on the demands of the context or situation.

Engagement States in Atypical Populations

Examination of engagement states has also focused on children with Down syndrome (DS) and ASD (Adamson, Bakeman, Deckner, & Romski, 2009; Lewy & Dawson, 1992). To be specific, Lewy and Dawson (1992) examined differences in engagement states in children with ASD and DS and typically developing children who all had similar levels of receptive language. These researchers found that children with ASD spent less time in CJE and more time in object engagement and being unengaged than did children with DS or typically developing children. In contrast, children with DS spent similar amounts of time in all engagement states as typically developing children (Lewy & Dawson, 1992).

Adamson et al. (2009) examined the developmental changes in JE over the course of 1 year in 30-month-old children with ASD and DS as compared with typically developing language-matched toddlers who had been studied longitudinally from 18 to 30 months. At 30 months, children with ASD spent more time in an unengaged state than did children with DS or typically developing 18-month-olds. Children with DS spent less time in an object state than the other two groups, and children with ASD spent more time in an object state. All three groups spent similar amounts of time in SJE, and SJE was the engagement state in which all groups spent the most time (i.e., about 50% of their time; Adamson et al., 2009). Last, children with ASD spent very little time (5%) in CJE as compared with the other two groups: 22% for children with DS and 16% for typically developing children (Adamson et al., 2009).

Taken together, these results suggest that the use of different engagement states varies on the basis of the child's etiology and development level (Adamson et al., 2009; Lewy & Dawson, 1992). In addition, these findings highlight how the behavioral phenotype associated with a disorder can influence engagement states (e.g., more object engagement in children with ASD and more CJE in children with DS). It is important to note that despite these different patterns of engagement, both children with DS and with ASD showed increasing SJE and CJE over time, similar to typically developing children (Adamson et al., 2009).

Engagement States and Language Development

Of the different domains that JE influences, language development may be particularly important because, as already noted, social engagement serves as a foundation for later language development (Bopp & Mirenda, 2011; Brady, Marquis, Fleming, & McLean, 2004; Smith, Adamson, & Bakeman, 1988; Thurm, Lord, Lee, & Newschaffer, 2007). Impairments in social engagement are a predictor of subsequent delays in expressive and receptive language, especially in children with ASD (Anderson et al., 2007; Thurm et al., 2007). In an examination of SJE, Bottema-Beutel, Yoder, Hochman, and Watson (2014) found that the use of what the researchers called *higher-order supported joint engagement*—defined as the child interacting more reciprocally with the social partner—significantly predicted social communication and expressive language in preschool children with ASD at an 8-month follow-up. In addition, early interventions targeting JE behaviors have indicated long-term effects for language outcomes for children with ASD (Kasari, Freeman, & Paparella, 2006, Kasari, Gulsrud, Freeman, Paparella, & Hellemann, 2012). It is possible that JE also serves as a predictor of subsequent language development and is a potential target for early intervention in children with FXS.

Early Social Development in FXS

Little research has been done examining early social behavior in FXS. However, other skills that support early social development are known areas of deficit in FXS. These skills include eye gaze (I. L. Cohen et al., 1991; Murphy, Abbeduto, Schroeder, & Serlin, 2007), especially for initiating and maintaining social interaction (Wolff, Gardner, Paccia, & Lappen, 1989), and dividing and switching attention between visual stimuli (Scerif, Cornish, Wilding, Driver, & Karmiloff-Smith, 2004). Further, it has been noted that individuals with FXS experience difficulties with social interactions across their life spans (Bailey et al., 1998).

Early Language Development in FXS

Most children with FXS experience delays in expressive and receptive language (Hatton et al., 2009; Mirrett, Bailey, Roberts, & Hatton, 2004; Roberts et al., 2009), but there have been a few studies examining the roots of these language deficits. These studies on early communication development suggest that evidence for delays in language can be traced to deficits in early social communication behaviors. For example, deficits in early communicative gesture use have been reported (Flenthrope & Brady, 2010) as well as deficits in joint attention behaviors (e.g., commenting on an object by vocalizing, using gestures, and/or eye gaze shifts; Marschik et al., 2014; Roberts, Mirrett, Anderson, Burchinal, & Neebe, 2002). These deficits are likely to affect SJE and CJE because gestures, vocalizations, and joint attention are frequent means by which children demonstrate JE.

Present Study

We examined the use of JE, both SJE and CJE, during toddlerhood (i.e., 24 to 36 months) in 28 young children with FXS. Because CJE is a more complex behavior than SJE, we also examined differences between these types of JE. Furthermore, we examined the relationship between JE, expressive and receptive language—using developmental, functional, and observational language measures—and level of ASD symptomatology in young children with FXS. Language and ASD symptomatology were measured at two time points in development, when children were between 24 and 36 months (i.e., toddler period) and later when these children were between 59 and 68 months (i.e., child period) in order to examine concurrent and predictive relationships between JE, language, and ASD symptomatology.

Research Questions

1. Do children with FXS between 24 and 36 months spend different amounts of time in SJE as opposed to CJE?
2. What is the nature of the relationship between early JE and expressive and receptive language ability during the toddler period and later in the child period?
3. What is the nature of the relationship between early JE and level of ASD symptomatology in FXS?

Method

Participants

Participants were 28 young children with FXS (24 boys, four girls) and their mothers who were recruited as part of a larger study on family adaptation to FXS (see Brady, Warren, Fleming, Keller, & Sterling, 2014, and Warren, Brady, Sterling, Fleming, & Marquis, 2010, for details). For this study, we focused on two time points in our data collection: 24 to 36 months, referred to as the *toddler period*, and 59 to 68 months, referred to as the *child period* (see Table 2 for developmental and descriptive information). At the toddler period, children with FXS had a mean chronological age of 33.18 months, and at the child period, children with FXS had a mean chronological age of 65.85 months. All of the mothers were premutation carriers. The median household income was $77,500 (range $32,000 to $400,000 measured between 2003 and 2005).

Procedure

These families were recruited across the United States from a national research registry, FXS family support groups, and from advertising at national conventions and on an FXS parent Listserv. Two trained graduate research assistants visited families in their own homes. During these visits, the Mullen Scales of Early Learning (MSEL; Mullen, 1995) was administered to children, and the Wechsler Adult Intelligence Scale–Third Edition (Wechsler, 1997) and Vineland Adaptive Behavior Scales (VABS; Sparrow, Balla, & Cicchetti, 1984; Sparrow, Cicchetti, & Balla, 2005) were administered to mothers. In addition, the mother–child dyads were videotaped interacting with one another in a series of structured and unstructured contexts (see description below). Immediately following the home visit, the graduate research assistants scored the Childhood Autism Rating Scale (CARS; Schopler et al., 1988) on the basis of what they had observed at the home visit.

Measures

JE

The coding scheme for JE was based on the coding scheme developed by Bakeman and Adamson (1984). The definitions of the five engagement states coded in this study are presented in Table 1. Numerous studies have used this coding scheme to examine JE between children and their mothers in typical and atypical populations (e.g., Adamson et al., 2004, 2009; Bottema-Beutel et al., 2014; Lewy & Dawson, 1992). JE was coded from a 5-min free play interaction context during the toddler period (i.e., 24 to 36 months). Mothers were asked to interact with their child as they normally would while playing with a selected set of developmentally appropriate toys (i.e., blocks, tower of rings, shapes).

The 5-min free play video was digitized and coded for JE using Noldus Observer software (Noldus Information Technology, 2002). An engagement state was identified as “a period of at least 3 seconds that is characterized by the child’s active interest in people and in objects and events” (Adamson et al., 2004, p. 1176). For each video, coders would watch the video in real time and code in Noldus when an engagement state started and ended. In order to identify the beginning and end of an engagement state, coders would look for break points (Bakeman & Adamson, 1984; Newtson, 1973) in the flow of the interaction between the mother and child. To ensure that an engagement state was started and stopped at the correct moment, after noting a change in state, coders would go back and rewatch the video until they felt they had pinpointed the most accurate break point. In addition, coders would check that the interaction lasted for at least 3 s to ensure that the dyad met the criteria for being in an engagement state.

Coding Reliability for JE

Two coders (graduate research assistants) who were naïve to the hypotheses of this study coded the video records. The first author trained coders on the coding scheme by explaining the scheme in depth and providing examples of the behaviors to be coded. Next, the first author and coders coded two practice videos not used in this study at the same time, discussing each coding decision. Coders then independently coded eight practice videos not used in this study until there was an 80% agreement between them. For the practice videos, if an 80% agreement was not achieved, they would compare coding and any disagreements would be resolved between them. If the disagreement could not be resolved, then the first author was asked to aid in clarifying the coding scheme. After training, each coder was randomly assigned 14 videos to code as the primary coder. Both coders coded 25% of videos, and reliability was calculated. Intraclass correlation coefficients (ICCs; Shrout & Fleiss, 1979) were calculated between the primary and reliability coder for the length of time the child spent in each engagement state (see Table 1 for definitions). For each state, the ICCs were unengaged .99, person .99, object .76, SJE .96, and CJE .98. Object engagement rarely occurred in the present study ($M = 0.04$, $SD = 0.05$), which may account for the lower ICC coefficient.

Table 2. Participant characteristics (*N* = 28).

Characteristic	Percentage	Toddler period			Child period		
		M	*SD*	Range	*M*	*SD*	Range
Child							
Age (in months)		33.18	0.36	25–36	65.86	2.55	59–68
Early learning composite		55.46	11.61	49–101	55.25	12.50	49–99
Mental age (in months)[a]		18.82	6.04	7.75–38.00	35.46	13.28	13.75–67.25
MSEL expressive language raw score		16.68	6.78	4–33	30.39	9.66	11–47
MSEL receptive language raw score		16.68	6.78	5–33	33.61	7.92	17–48
VABS expressive language raw score		14.21	7.69	6–39	32.25	13.95	10–58
VABS receptive language raw score		17.50	3.34	13–24	23.07	1.78	19–26
Rate of different words		1.18	1.58	0.00–5.68	4.17	2.63	0.00–9.69
ASD symptomatology[b]		25.77	6.20	15.5–38.0	25.59	6.33	16–39
Ethnicity (%)							
Caucasian	96.4						
Hispanic	3.6						
Maternal							
Age (in years)		32.87	4.97	20.50–41.75			
IQ		110.43	13.87	73–130			
Education (%)							
Some high school	3.6						
High school graduate	14.2						
Some college	21.5						
College graduate	32.2						
Postgraduate training	28.6						

Note. MSEL = Mullen Scales of Early Learning; VABS = Vineland Adaptive Behavior Scales; ASD = autism spectrum disorder.

[a]Overall mental age calculated from the age equivalence scores for the fine motor, visual reception, expressive language, and receptive language domains of the MSEL. [b]Childhood Autism Rating Scale score.

MSEL

The MSEL is a standardized assessment for children between the ages of 0 and 68 months (Mullen, 1995). The MSEL assesses five domains of development: gross motor, fine motor, visual reception, expressive language, and receptive language. *T* scores are calculated for the five domains and then combined to create an overall standard score that reflects an estimate of overall development (i.e., early learning composite). We used raw scores for expressive and receptive language to describe the child's language at the toddler and child periods. The MSEL has well-established construct validity, content validity, predictive validity, and strong concurrent validity with other developmental tests for children (e.g., Bayley Scales of Infant Development [Bayley, 1993]; Mullen, 1995).

VABS–Interview Edition, Survey Form

The VABS is a standardized parent interview designed to examine the personal and social functioning of individuals from birth through adulthood (Sparrow et al., 1984, 2005). For the present study, only the receptive and expressive communication subdomains were examined at each time point. VABS items are scored using a 3-point Likert scale (*never*, *sometimes/partially*, or *usually*). Items may also be scored as "parent does not know if the child can perform the activity" or as "the child has not had the opportunity to perform the activity." The VABS has established construct, criterion, and content validity and has satisfactory levels of internal consistency.

Rate of Different Words per Minute

As part of the larger study (Brady et al., 2014), each child's utterances produced while interacting with his or her mother during four contexts—joint book reading, having a snack, free play, and unstructured daily activities—were transcribed from the video records and analyzed using the Systematic Analysis of Language Transcriptions (Miller & Chapman, 1985). A total of 25 min of interaction from these contexts was used for transcription (i.e., 5 min each from book reading, snack, and free play as well as 10 min of daily activities at both the toddler and child period). The average length of these interactions was 24.8 min (*SD* = 1.21 min). Because the lengths of these observations varied slightly, we calculated the child's rate of different words per minute at each time point (i.e., the toddler and child periods) by adding all the different words produced during the language sample and dividing by the total time of each of the language samples.

Coding Reliability for Rate of Different Words per Minute

Research assistants completed the primary and reliability transcribing for the number of different words. Each transcriber was trained to 80% reliability with the second author on a set of videos not used in this study. Following this training, each transcriber independently transcribed the video records and compared transcripts for agreement. If their agreement was below 80%, they resolved their differences by consensus. The total number of different words identified by each transcriber was used in reliability calculations rather than a word-by-word comparison. ICCs between the primary and reliability transcriber were .97.

CARS

The CARS is a 15-item measure of general autistic behavior (Schopler et al., 1988) that is scored on a 4-point Likert scale (*normal/age appropriate*, *mildly abnormal*, *moderately abnormal*, and *severely abnormal*). The specific symptoms rated on the CARS are relating to people; object use; imitation; emotional response; body use; object use; adaptation to change; visual response; listening response; taste, smell, and touch responses and use; fear or nervousness; verbal communication; nonverbal communication; and activity level. In addition, the general impressions (i.e., absence or presence of ASD) are rated. The total score of the 15 items provides a continuum of autistic behavior that can also be interpreted as a performance measure (Schopler et al., 1988). Using this method, a score below 30 indicates low or no autistic behaviors, a score from 30 to 36 indicates mild-to-moderate autistic behavior, and a score above 36 indicates high autistic behavior (Schopler et al., 1988). Graduate research assistants were trained on the CARS scoring using the provided training tapes. Scoring was done by consensus immediately following the home observation.

Data Analysis Plan

To examine our first research question about whether there were differences in the amount of time spent in SJE and CJE, we used paired samples t tests. For this analysis, the proportion of time spent in each engagement state was used instead of length of time because there was some variation in the length of the free play. The average length of the free play was about 5 min (i.e., 300 s; $M = 299.17$, $SD = 6.99$, range 285.75–325.51).

We used Pearson r correlations to address our second and third research questions about the nature of the relationship between JE, language abilities at two time periods, and ASD symptomatology at the two time periods. Although our sample size was small, scores were normally distributed; therefore, Pearson r correlations could be used rather than a nonparametric statistic, such as Spearman's rho. To be specific, for our second research question, we used partial correlations controlling for chronological age in the toddler period when JE was measured to account for the effect of age on language at both the toddler and child periods. For these analyses, the raw scores from the MSEL and VABS were used because the standard scores for these variables had floor effects with more than 50% of the sample scoring uniformly at the lowest possible score. For our third research question, we examined the relationship of JE in the toddler period to ASD symptomatology at both the toddler and child periods but did not control for chronological age in the toddler period.

We used a weighted JE score to indicate each child's level of JE development. At first, we attempted to examine the associations between proportion of time spent in both SJE and CJE and other variables; however, drawing inferences about these correlations proved to be problematic. For the SJE correlations, the same SJE score represented very different profiles of JE behavior (see online Supplemental Table 1 for the distributions of these variables). For example, participant 11 spent no time in SJE because 87% of the time was spent in CJE. Participant 17 also spent no time in SJE but also spent no time in CJE because time was spent unengaged or in person engagement. Correlations with CJE were problematic because many participants did not use CJE, and thus there was restriction in the range. Using a simple summed JE score (i.e., SJE + CJE = total JE) did not account for the important distinctions between SJE and CJE as proposed by Adamson and Bakeman (1991), and a large percentage of the sample would have scores that could not be used in analysis because they would add to the maximum scores of 1.00. As a consequence, we created a composite variable with a distribution suitable for analysis and reflective of CJE being relatively more complex than SJE.

We weighted the proportion of time spent in CJE more highly than SJE because it is a more complex behavior that involves dynamic and reciprocal alternating of attention and sharing of interest between the object and the caregiver (Adamson et al., 2004; Bakeman & Adamson, 1984). In addition, because there are differences in the developmental timing of these behaviors (i.e., SJE is used consistently after 6 months, and CJE is used consistently after 13 months), weighting CJE is more representative of the developmental gains associated with SJE and CJE. Similar weighting procedures have been used to reflect growth in early communication skills (e.g., Greenwood, Ward, & Luze, 2003; Luze et al., 2001) and complexity of play behaviors (Thiemann-Bourque, Brady, & Fleming, 2012). Similar to these other studies, we rank-ordered behavior from least to most complex. SJE was ranked as 1 = *less complex*, and CJE was ranked as 2 = *more complex*—therefore, we multiplied CJE by two. Thus, a weighted JE score was calculated by taking the CJE score times two and adding the SJE score for each child. For example, if proportion of time in CJE was .25 and proportion of time in SJE was .40, then the weighted JE score would be 0.90 (i.e., $[.25 \times 2] + .40 = .90$). Weighted JE scores ranged from 0.00 to 2.00 with a mean of 1.48 ($SD = 0.36$) and were more normally distributed, which helped to address issues of restriction in the range (see online Supplemental Table 1). This weighted JE score was used for all correlational analyses.

Results

Differences in SJE and CJE

To characterize the pattern of engagement states in children with FXS, we examined the mean amount of time children spent in each engagement state during interactions with their mothers during toddlerhood (i.e., 24 to 36 months; see Table 3). Children spent, on average, the most time in SJE, followed by CJE. A paired samples t test indicated that they spent significantly more time in SJE than CJE,

Table 3. Frequency and mean proportion of time spent in each engagement state ($N = 28$).

Engagement State	Number of children who engaged in state	Mean proportion of time	*SD*	Range
Unengaged	2	.02	.11	0.00–.59
Person	2	.04	.16	0.00–.83
Object	11	.04	.05	0.00–.15
Supported joint engagement	25	.76	.31	0.00–1.00
Coordinated joint engagement	12	.15	.26	0.00–1.00

$t(27) = 6.03$, $p < .001$, $d = 1.15$. However, as Table 3 indicates, profiles were extremely variable for each engagement state. Because of the potential influence of outliers on the mean difference test, a Wilcoxon signed-ranks test was also conducted, which also indicated children spent more time in SJE than in CJE, $z = 3.69$, $p = .001$. Only two children spent more time in CJE than in SJE (mean rank = 18), and 25 children spent less time in CJE than in SJE (mean rank = 13.68). One child spent an equal amount of time in SJE and CJE. Children spent a small amount of time in object engagement and even less time in person or unengaged states.

Weighted JE and Language Abilities

Partial correlations were performed to examine the nature of the relationship between weighted JE, measured in the toddler period, and language (i.e., the receptive and expressive scales from the MSEL and the VABS and rate of different words per minute) measured at both the toddler and child periods controlling for chronological age in the toddler period.

Toddler Period

For the toddler period, children who had higher weighted JE also had a higher rate of different words per minute, $r(28) = .66$, $p < .001$, and higher expressive language as measured by the VABS, $r(28) = .40$, $p = .04$, when controlling for chronological age in the toddler period (see online Supplemental Table 2 for full correlation matrix). There were no other statistically significant relationships during the toddler period.

Child Period

Children who had higher weighted JE during the toddler period had higher expressive and receptive language raw scores on both the MSEL, expressive: $r(28) = .51$, $p = .007$; receptive: $r(28) = .49$, $p = .009$, and on the VABS, expressive: $r(28) = .49$, $p = .009$; receptive: $r(28) = .42$, $p = .03$, during the child period when controlling for chronological age in the toddler period (see online Supplemental Table 3 for full correlation matrix). In addition, the partial correlations indicated that children who had higher weighted JE during the toddler period had higher rates of different words per minute in the child period when controlling for chronological age in the toddler period, $r(28) = .39$, $p = .047$.

Weighted JE and Level of ASD Symptomatology

Pearson *r* correlations were used to examine the nature of the relationship between weighted JE, measured during the toddler period, and ASD symptomatology at both the toddler and child periods. Results indicate that during the toddler period, children who had a higher level of ASD symptomatology had lower weighted JE scores, $r(28) = -.52$, $p = .005$. The relationship between weighted JE in the toddler period and level of ASD symptomatology in the child period was not significant, $r(28) = -.34$, $p = .08$; however, the directions of effects for the child period was the same as the concurrent association in the toddler period.

Comparison of Those Who Did and Did Not Use CJE

Following our analysis, an additional question arose: Are there differences between the 12 participants who used CJE and those who never used CJE? To examine this question, independent samples *t* tests were performed to examine differences in overall development (as measured by the early learning composite from the MSEL), language (i.e., expressive and receptive language as measured by the MSEL and VABS and rate of different words per minute), and ASD symptomatology at both the toddler and child periods between those who did use CJE and those who did not use CJE. Results indicated that those who used CJE as a toddler tended to be more advanced developmentally and to use more words at both the toddler and child periods. Although these differences were not statistically significant, in the toddler period, the effect size for differences between those who used and did not use CJE were large for the early learning composite scores, $t(11.48) = 1.87$, $p = .088$, $d = .93$, and for rate of different words per minute, $t(13.85) = 2.15$, $p = .05$, $d =.94$ (J. Cohen, 1988). In the child period, the effect size for differences in early learning composite scores was also large, $t(11.87) = 1.98$, $p = .07$, $d = .95$, and the effect size for differences in rate of different words per minute was moderate, $t(26) = 1.48$, $p = .15$, $d = .56$ (J. Cohen, 1988; see online Supplemental Figures 1 and 2 for distributions of these variables).

Discussion

The present study sought to characterize JE in young children with FXS and the relationship of these behaviors to language development and the extent of ASD symptoms.

To our knowledge, this is the first study to examine these behaviors in FXS. Our results indicate that during the toddler period, children with FXS spent significantly more time in SJE versus CJE. This pattern of more time spent in SJE versus CJE is similar to the patterns that have previously been observed in typically developing 12- to 15-month-old infants (Bakeman & Adamson, 1984) and typically developing 18-month-olds (Adamson et al., 2009). Furthermore, previous research found that children with DS and ASD at 30 months of age also spent most of their time in SJE—a pattern similar to typically developing 18-month-olds (Adamson et al., 2009). The mean mental age for our sample when JE was examined was 18.82 months, as measured by the MSEL, with about half of the sample having a mental age at this level or younger. Therefore, it appears that children with FXS are showing JE behaviors in the toddler period that are commensurate with their overall developmental level (Murphy & Abbeduto, 2005).

Our results also provide preliminary evidence that children with FXS who used CJE in the toddler period tend to be more developmentally advanced and use more words both concurrently (i.e., toddler period) and later in development (i.e., child period). However, although the effect sizes for these differences were moderate to large, these effects were not statistically significant, most likely because of the small sample size of the present study (J. Cohen, 1988). More research is needed to further elucidate the effect of developmental level on JE in FXS.

In regards to severity of impairment, we found that during the toddler period, children with FXS who had more ASD symptoms also had lower weighted JE scores. The same direction of effects was observed between weighted JE scores in the toddler period and later ASD symptomology, but this relationship did not reach statistical significance. It is possible that the relationship between JE and ASD symptomatology is weaker when the two variables are measured at different points in time, such that if JE was also measured at the child period, the relationship might reach statistical significance. Nonetheless, this direction of effects is commensurate with past research indicating that joint attention—which is similar to JE—is negatively related to ASD symptoms (Mundy & Vaughan, 2002) and that children with ASD spend less time in CJE than typically developing children or children with DS (Adamson et al., 2009; Lewy & Dawson, 1992). Thus, it seems that the use of different types of JE are important to consider when evaluating ASD symptoms in FXS. More research is needed to evaluate the clinical implications of JE for early identification of ASD in FXS.

In the present study, we examined language holistically by including measures of developmental, functional, and observed language at two time points (i.e., the toddler and child periods). Examination of language in the toddler period indicated that those who had higher weighted JE scores also had higher observed expressive language (i.e., rate of different words per minute in a language sample) and functional expressive language (i.e., the VABS) when controlling for chronological age in the toddler period. Considering that JE involves behaviors related to prelinguistic communication (i.e., eye gaze, gestures, etc.; Bates, Benigni, Bretherton, Camaioni, & Volterra, 1979), it seems that the ability to use JE also helps to foster expressive communication. Furthermore, weighted JE continued to be related to later expressive language as measured by all three of our language measures when controlling for chronological age during the toddler period. Thus, relationships between later expressive language and JE were observed in children with FXS similar to those that have been reported for typically developing children and children with other IDDs, including ASD (e.g., Bopp & Mirenda, 2011; Brady et al., 2004; Smith et al., 1988). Taken together, it appears that over the course of early development, the use of JE is related to more advanced expressive language at both the toddler and child periods for children with FXS.

Our findings are consistent with Adamson and Bakeman's view that dynamic sharing of interest between an object and caregiver promotes higher expressive language (Adamson et al., 2004; Bakeman & Adamson, 1984). Further, this study adds support to Bottema-Beutel et al.'s (2014) hypothesis that more reciprocal and collaborative forms of JE, such as our weighted JE variable, are especially important for promoting social communication and expressive language. Interventions that also target social sharing and social interaction, especially through parent scaffolding, may help to promote language development by helping children transition into more complex forms of JE.

In regards to receptive language, those who had higher weighted JE scores in the toddler period also had higher developmental and functional receptive language scores (i.e., the MSEL and VABS, respectively) during the child period when controlling for chronological age in the toddler period. These findings are consistent with earlier findings showing that JE provides opportunities for additional verbal input when the child and mother are focused on the same activity (e.g., Mundy, Kasari, Sigman, & Ruskin, 1995). When this is the case, vocabulary uptake may be facilitated (McDuffie & Yoder, 2010). Future studies on JE in children with FXS might explore the relationship between object engagement and receptive language as other studies have indicated a positive relationship (Bottema-Beutel et al., 2014). Child object engagement was observed sparingly in our study (mean proportion of time spent in object engagement = .04); therefore, we were unable to explore this relationship.

Limitations of the Current Study

We had a small sample of JE behavior—only a 5-min free play—compared with some studies. Although numerous other studies of parent–child interactions also used samples of behavior that range from 2 to 5 min (e.g., Meek, Robinson, & Jahromi, 2012; Siller, Hutman, & Sigman, 2013) or 6 to 10 min (e.g., Patterson, Elder, Gulsrud, & Kasari, 2014; Wan et al., 2013), 5 min of observation time is short in comparison with other studies that observed parent–child interactions from 15 to 40 min comprised of playing with toys, making music, book reading, etc. (Adamson et al., 2004; Bottema-Beutel et al., 2014). Although there is no standard length of time for examining JE during parent–child interactions, it is possible that the length of our observation may have skewed our findings. For example, it is possible that mothers were more successful at maintaining an interaction with their child for 5 min—leading to periods of SJE and CJE—but if observed over a longer interval of time, more time spent in object engagement or lack of engagement would have been observed.

Also, one of the main goals in the larger study (Brady et al., 2014; Warren et al., 2010) was to examine parenting in mothers of children with FXS. The mothers' knowledge of our interest in parenting may have influenced their interaction style such that they included more parental scaffolding, which thereby facilitated more opportunities for SJE with their children. However, mothers were not part of any intervention aimed at changing their parenting behaviors, so it is likely that the parents' behavior was fairly typical.

There were additional limitations associated with procedures used in this study. We did not have a comparison group of developmentally matched typically developing children or children with IDD. Including developmentally matched comparison groups in future studies could allow for a determination of whether JE is simply delayed in FXS or if there is a syndrome-specific pattern of development of JE in FXS. The current sample had only four girls with FXS. Considering that girls with FXS are less affected than boys (Hagerman, 2007), future studies with a larger sample of girls should explore sex differences in JE. Last, although the use of the weighted JE variable allowed us to investigate JE within this limited time sample, future investigations may wish to examine CJE separately from SJE in order to examine relative contributions of each type of JE to language development and ASD symptomology.

Conclusions and Implications

JE behaviors are some of the earliest social and language skills that can be examined. Differences in the development of JE behaviors in FXS may have lifelong implications for all domains of development but especially for the domains of language development, social communication, and social interaction—areas of known deficits in FXS. In addition, early impairments in JE behaviors may serve as an early indicator of ASD in children with FXS (Mundy & Vaughan, 2002). Promoting growth and development of JE behaviors through early intervention may be a pivotal focus during the key window for early intervention—birth to 5 years—and influence multiple domains of development.

Existing interventions that target JE and other early prelinguistic communication skills in children with IDD and language delays have shown long-lasting effects in terms of language and social engagement (Fey et al., 2013; Girolametto & Weitzman, 2006; Kasari et al., 2012; Landry, Smith, Swank, & Guttentag, 2008). These existing interventions may need modifications to better fit the needs of children with FXS by taking into consideration the behavior phenotype associated with FXS (e.g., account for how the presence of ASD or other challenging behaviors associated with FXS may affect response to intervention). In a similar manner, most of these interventions are designed for children who are prelinguistic and, therefore, may need to be slightly adjusted to be more developmentally appropriate for prelinguistic infants. Last, including a parent component —as many of these existing early interventions do—is key as parents can be taught to help support and promote these behaviors in infants and young children during day-to-day interactions.

There are many new avenues for research on JE, joint attention, and social communication in FXS. These range from the basic tasks of better understanding the development and patterns of strengths and weakness of these behaviors to translating research into effective interventions. An especially important aspect of this research involves collaborations between researchers and clinicians to help modify and implement effective intervention strategies to promote later language, social communication, and social interaction in individuals with FXS. The present study adds to the growing literature on identifying profiles of early social and communication behaviors in FXS (Flenthrope & Brady, 2010; Marschik et al., 2014; Roberts et al., 2002). To be specific, our findings highlight the need to more closely examine the developmental course of JE and early social behaviors in children with FXS, starting in infancy when these behaviors emerge.

Acknowledgments

This research was supported by National Institute on Child Health and Human Development Grants T32 HD057844 (awarded to Kathryn J. Saunders), P30 HD02528 (awarded to John A. Colombo), and P30 HD003110 (awarded to Steven F. Warren). This manuscript was presented as a poster at the proceedings of the 47th Annual Gatlinburg Conference on Research and Theory in Intellectual and Developmental Disabilities, Chicago, Illinois, and at the proceedings of the Second Annual KU Postdoc Research Day, Lawrence, Kansas. We would like to thank Carrissa Phillippe and Alisa Miller, who assisted in data coding.

References

Abbeduto, L., Brady, N., & Kover, S. T. (2007). Language development and fragile X syndrome: Profiles, syndrome-specificity, and within-syndrome differences. *Mental Retardation and Developmental Disabilities Research Reviews, 13,* 36–46. doi:10.1002/mrdd

Abbeduto, L., Evans, J., & Dolan, T. (2001). Theoretical perspectives on language and communication problems in mental retardation and developmental disabilities. *Mental Retardation and Developmental Disabilities Research Reviews, 7,* 45–55.

Adamson, L. B., & Bakeman, R. (1991). The development of shared attention during infancy. In R. Vasts (Ed.), *Annals of child development* (Vol. 8, pp. 1–41). London, England: Jessica Kingsley.

Adamson, L. B., Bakeman, R., & Deckner, D. F. (2004). The development of symbol-infused joint engagement. *Child Development, 75,* 1171–1187. doi:10.1111/j.1467-8624.2004.00732.x

Adamson, L. B., Bakeman, R., Deckner, D. F., & Romski, M. (2009). Joint engagement and the emergence of language in children with autism and Down syndrome. *Journal of Autism and Developmental Disorders, 39,* 84–96. doi:10.1007/s10803-008-0601-7

Adamson, L. B., & Chance, S. E. (1998). Coordinating attention to people, objects, and language. In A. M. Wetherby, S. F. Warren, & J. Reichle (Eds.), *Transitions in prelinguistic communication* (Vol. 7, pp. 15–37). Baltimore, MD: Brookes.

Anderson, D. K., Lord, C., Risi, S., DiLavore, P. S., Shulman, C., Thurm, A., . . . Pickles, A. (2007). Patterns of growth in verbal abilities among children with autism spectrum disorder. *Journal of Consulting and Clinical Psychology, 75*(4), 594–604. doi:10.1037/0022-006X.75.4.594

Bailey, D. B., Jr., Mesibov, G. B., Hatton, D. D., Clark, R., Roberts, J. E., & Mayhew, L. (1998). Autistic behavior in young boys with fragile X syndrome. *Journal of Autism and Developmental Disorders, 28,* 499–508. doi:10.1023/A:1026048027397

Bailey, D. B., Jr., Raspa, M., Olmsted, M., & Holiday, D. B. (2008). Co-occurring conditions associated with FMR1 gene variations: Findings from a national parent survey. *American Journal of Medical Genetics Part A, 146A,* 2060–2069. doi:10.1002/ajmg.a.32439

Bakeman, R., & Adamson, L. B. (1984). Coordinating attention to people and objects in mother-infant and peer-infant interaction. *Child Development, 55,* 1278–1289.

Bates, E., Benigni, L., Bretherton, I., Camaioni, L., & Volterra, V. (1979). *The emergence of symbols: Cognition and communication in infancy*. New York, NY: Academic Press.

Bayley, N. (1993). *Bayley Scales of Infant Development–Second Edition*. San Antonio, TX: The Psychological Corporation.

Bopp, K. D., & Mirenda, P. (2011). Prelinguistic predictors of language development in children with autism spectrum disorders over four-five years. *Journal of Child Language, 38,* 1–19. doi:10.1017/S0305000910000140

Bottema-Beutel, K., Yoder, P. J., Hochman, J. M., & Watson, L. R. (2014). The role of supported joint engagement and parent utterances in language and social communication development in children with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 44,* 2162–2174. doi:10.1007/s10803-014-2092-z

Brady, N. C., Marquis, J., Fleming, K., & McLean, L. (2004). Prelinguistic predictors of language growth in children with developmental disabilities. *Journal of Speech, Language, and Hearing Research, 47,* 663–677.

Brady, N., Warren, S. F., Fleming, K., Keller, J., & Sterling, A. (2014). Effect of sustained maternal responsivity on later vocabulary development in children with fragile X syndrome. *Journal of Speech, Language, and Hearing Research, 57,* 212–226. doi:10.1044/1092-4388(2013/12-0341)212

Centers for Disease Control and Prevention. (2011). *Facts about fragile X syndrome*. Retrieved from http://www.cdc.gov/ncbddd/actearly/pdf/parents_pdfs/fragile_x.pdf

Clifford, S., Dissanayake, C., Bui, Q. M., Huggins, R., Taylor, A. K., & Loesch, D. Z. (2007). Autism spectrum phenotype in males and females with fragile X full mutation and premutation. *Journal of Autism and Developmental Disorders, 37,* 738–747. doi:10.1007/s10803-006-0205-z

Cohen, I. L., Sudhalter, V., Pfadt, A., Jenkins, E. C., Brown, W. T., & Vietze, R. M. (1991). Why are autism and the fragile X syndrome associated? Conceptual and methodological issues. *American Journal of Human Genetics, 48,* 195–202.

Cohen, J. (1988). *Statistical power analysis for the behavioral sciences* (2nd ed.). Mahwah, NJ: Erlbaum.

Crawford, D. C., Acuna, J. M., & Sherman, S. L. (2001). FMR1 and the fragile X syndrome: Human genome epidemiology review. *Genetics in Medicine, 3,* 359–371.

Fey, M. E., Yoder, P. J., Warren, S. F., Bredin-Oja, S. L., Oetting, J., & Crais, E. (2013). Is more better? Milieu communication teaching in toddlers with intellectual disabilities. *Journal of Speech, Language, and Hearing Research, 56,* 679–693. doi:10.1044/1092-4388(2012/12-0061)

Fidler, D. J., Lunkenheimer, E., & Hahn, L. (2011). Emerging behavioral phenotypes and dynamic systems theory. *International Review of Research in Intellectual Disabilities, 40,* 17–42.

Flenthrope, J. L., & Brady, N. C. (2010). Relationships between early gestures and later language in children with fragile X syndrome. *American Journal of Speech-Language Pathology, 19,* 135–142. doi:10.1044/1058-0360(2009/09-0018)

Girolametto, L., & Weitzman, E. (2006). It takes two to talk: The Hanen program for parents: Early language intervention through caregiver training. In S. F. Warren & M. E. Fey (Series Eds.) & R. J. McCauley & M. E. Fey (Vol. Eds.), *Communication and language intervention series: Treatment of language disorders in children* (pp. 77–104). Baltimore, MD: Brookes.

Greenwood, C. R., Ward, S. M., & Luze, G. J. (2003). The early communication indicator (ECI) for infants and toddlers: What is it, where it's been, and where it needs to go. *The Behavior Analyst Today, 3,* 383–388.

Hagerman, R. J. (2007). Etiology, diagnosis, and development in fragile X syndrome. In J. E. Roberts, R. S. Chapman, & S. F. Warren (Eds.), *Speech and language development and intervention in Down syndrome and fragile X syndrome* (pp. 27–42). Baltimore, MD: Brookes.

Hagerman, R. J., Rivera, S. M., & Hagerman, P. J. (2008). The fragile X family of disorders: A model for autism and targeted treatments. *Current Pediatric Reviews, 4,* 40–52.

Hall, S. S., Lightbody, A. A., Hirt, M., Rezvani, A., & Reiss, A. L. (2010). Autism in fragile X syndrome: A category mistake? *Journal of the American Academy of Child & Adolescent Psychiatry, 49,* 921–933. doi:10.1016/j.jaac.2010.07.001

Hall, S. S., Lightbody, A. A., & Reiss, A. L. (2008). Compulsive, self-injurious, and autistic behavior in children and adolescents with fragile X syndrome. *American Journal on Mental Retardation, 113,* 44–53. doi:10.1352/0895-8017(2008)113[44:CSAABI]2.0.CO;2

Harris, S. W., Hessl, D., Goodlin-Jones, B., Ferranti, J., Bacalman, S., Barbato, I., . . . Hagerman, R. J. (2008). Autism profiles of males with fragile X syndrome. *American Journal on Mental Retardation, 113,* 427–438. doi: 10.1352/2008.113:427-438

Hatton, D. D., Wheeler, A., Sideris, J., Sullivan, K., Reichardt, A., Roberts, J., . . . Abbeduto, L. (2009). Developmental trajectories of young girls with fragile X syndrome. *American Journal of Intellectual and Developmental Disabilities, 114,* 161–171. doi:10.1352/1944-7558-114.3.161

Kasari, C., Freeman, S., & Paparella, T. (2006). Joint attention and symbolic play in young children with autism: A randomized controlled intervention study. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 47,* 611–620. doi:10.1111/j.1469-7610.2005.01567.x

Kasari, C., Gulsrud, A., Freeman, S., Paparella, T., & Hellemann, G. (2012). Longitudinal follow-up of children with autism receiving targeted interventions on joint attention and play. *Journal of the American Academy of Child & Adolescent Psychiatry, 51,* 487–495. doi:10.1016/j.jaac.2012.02.019

Kaufmann, W. E., Cortell, R., Kau, A. S. M., Bukelis, I., Tierney, E., Gray, R. M., . . . Stanard, P. (2004). Autism spectrum disorder in fragile X syndrome: Communication, social interaction, and specific behaviors. *American Journal of Medical Genetics Part A, 129A,* 225–234. doi:10.1002/ajmg.a.30229

Landry, S., Smith, K., Swank, P. R., & Guttentag, C. (2008). A responsive parenting intervention: The optimal timing across early childhood for impacting maternal behaviors and child outcomes. *Developmental Psychology, 44,* 1335–1353. doi:10.1037/a0013030

Lewy, A. L., & Dawson, G. (1992). Social stimulation and joint attention in young autistic children. *Journal of Abnormal Child Psychology, 20,* 555–566. doi:10.1007/BF00911240

Loesch, D. Z., Huggins, R. M., & Hagerman, R. J. (2004). Phenotypic variation and FMRP levels in fragile X. *Mental Retardation and Developmental Disabilities Research Reviews, 10,* 31–41. doi:10.1002/mrdd.20006

Luze, G. J., Linebarger, D. L., Greenwood, C. R., Carta, J. J., Walker, D., Leitschuh, C., & Atwater, J. B. (2001). Developing a general outcome measure of growth in the expressive communication of infants and toddlers. *School Psychology Review, 30,* 383–406.

Marschik, P. B., Bartl-Pokorny, K. D., Sigafoos, J., Urlesberger, L., Pokorny, F., Didden, R., . . . Kaufmann, W. E. (2014). Development of socio-communicative skills in 9- to 12-month-old individuals with fragile X syndrome. *Research in Developmental Disabilities, 35,* 597–602. doi:10.1016/j.ridd.2014.01.004

McDuffie, A., & Yoder, P. (2010). Types of parent verbal responsiveness that predict language in young children with autism spectrum disorder. *Journal of Speech, Language, and Hearing Research, 53,* 1026–1039. doi:10.1044/1092-4388(2009/09-0023)

Meek, S. E., Robinson, L. T., & Jahromi, L. B. (2012). Parent–child predictors of social competence with peers in children with and without autism. *Research in Autism Spectrum Disorders, 6,* 815–823.

Miller, J. F., & Chapman, R. S. (1985). *Systematic Analysis of Language Transcripts*. Madison, WI: University of Wisconsin–Madison, Waisman Center, Language Analysis Laboratory.

Mirrett, P. L., Bailey, D. B., Jr., Roberts, J. E., & Hatton, D. D. (2004). Developmental screening and detection of developmental delays in infants and toddlers with fragile X syndrome. *Journal of Developmental & Behavioral Pediatrics, 25,* 21–27.

Mullen, E. (1995). *Mullen Scales of Early Learning*. Circle Pines, MN: AGS.

Mundy, P., Kasari, C., Sigman, M., & Ruskin, E. (1995). Nonverbal communication and early language acquisition in children with Down syndrome and in normally developing children. *Journal of Speech and Hearing Research, 38,* 157–167.

Mundy, P., & Vaughan, A. (2002). Joint attention and its role in the diagnostic assessment of children with autism. *Assessment for Effective Intervention, 27,* 57–60. doi:10.1177/073724770202700108

Murphy, M., & Abbeduto, L. (2005). Indirect genetic effects and the early language development of children with genetic mental retardation syndromes: The role of joint attention. *Infants & Young Children, 18,* 47–59. doi:10.1097/00001163-200501000-00006

Murphy, M., Abbeduto, L., Schroeder, S., & Serlin, R. (2007). Contribution of social and information-processing factors to eye-gaze avoidance in fragile X syndrome. *American Journal on Mental Retardation, 112,* 349–360.

Newtson, D. (1973). Attribution and the unit of perception of ongoing behavior. *Journal of Personality and Social Psychology, 28,* 28–38.

Noldus Information Technology. (2002). *The observer, base package for windows reference manual 5th edition*. Wageningen, the Netherlands: Author.

Patterson, S. Y., Elder, L., Gulsrud, A., & Kasari, C. (2014). The association between parental interaction style and children's joint engagement in families with toddlers with autism. *Autism, 18,* 511–518.

Roberts, J. E., Mankowski, J. B., Sideris, J., Goldman, B. D., Hatton, D. D., Mirrett, P. L., . . . Bailey, D. B., Jr. (2009). Trajectories and predictors of the development of very young boys with fragile X syndrome. *Journal of Pediatric Psychology, 34,* 827–836. doi:10.1093/jpepsy/jsn129

Roberts, J. E., Mirrett, P., Anderson, K., Burchinal, M., & Neebe, E. (2002). Early communication, symbolic behavior, and social profiles of young males with fragile X syndrome. *American Journal of Speech-Language Pathology, 11,* 295–304. doi:10.1044/1058-0360(2002/034)

Scerif, G., Cornish, K., Wilding, J., Driver, J., & Karmiloff-Smith, A. (2004). Visual search in typically developing toddlers and toddlers with fragile X or Williams syndrome. *Developmental Science, 7,* 116–130. doi:10.1111/j.1467-7687.2004.00327.x

Schopler, E., Reichler, R. J., & Renner, B. R. (1988). *The Childhood Autism Rating Scale (CARS)*. Los Angeles CA: Western Psychological Services.

Shrout, P. E., & Fleiss, J. L. (1979). Intraclass correlations: Uses in assessing rater reliability. *Psychological Bulletin, 2,* 420–428.

Siller, M., Hutman, T., & Sigman, M. (2013). A parent-mediated intervention to increase responsive parental behaviors and child communication in children with ASD: A randomized clinical trial. *Journal of Autism and Developmental Disorders, 43,* 540–555.

Smith, C. B., Adamson, L. B., & Bakeman, R. (1988). Interactional predictors of early language. *First Language, 8,* 143–156. doi:10.1177/014272378800802304

Sparrow, S. S., Balla, D. A., & Cicchetti, D. (1984). *Vineland Adaptive Behavior Scales*. Circle Pines, MN: AGS.

Sparrow, S. S., Cicchetti, D., & Balla, D. A. (2005). *Vineland-II: Vineland Adaptive Behavior Scales–Second Edition*. Minneapolis, MN: Pearson.

Thiemann-Bourque, K. S., Brady, N. C., & Fleming, K. K. (2012). Symbolic play of preschoolers with severe communication impairments with autism and other developmental delays: More similarities than differences. *Journal of Autism and Developmental Disorders, 42,* 863–873. doi:10.1007/s10803-011-1317-7

Thurm, A., Lord, C., Lee, L. C., & Newschaffer, C. (2007). Predictors of language acquisition in preschool children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 37,* 1721–1734.

Trevarthen, C., & Aitken, K. J. (2001). Infant intersubjectivity: Research, theory, and clinical applications. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 42,* 3–48.

Trevarthen, C., & Hubley, P. (1978). Secondary intersubjectivity: Confidence, confiding and acts of meaning in the first year. In A. Lock (Ed.), *Action, gesture and symbol* (pp. 183–229). London, England: Academic Press.

Verkerk, A. J. M. H., Pieretti, M., Sutcliffe, J. S., Fu, Y.-H., Kuhl, D. P. A., Pizzuti, A., . . . Warren, S. T. (1991). Identification of a gene (FMR-1) containing a CGG repeat coincident with a breakpoint cluster region exhibiting length variation in fragile X syndrome. *Cell, 65,* 905–914. doi:10.1016/0092-8674(91)90397-H

Wan, M. W., Green, J., Elsabbagh, M., Johnson, M., Charman, T., & Plummer, F. (2013). Quality of interaction between at-risk infants and caregiver at 12–15 months is associated with 3-year autism outcome. *The Journal of Child Psychology and Psychiatry, 54,* 763–771.

Warren, S., Brady, N., Sterling, A., Fleming, K., & Marquis, J. (2010). Maternal responsivity predicts language development in young children with fragile X syndrome. *American Journal on Intellectual and Developmental Disabilities, 115,* 54–75. doi:10.1352/1944-7558-115.1.54

Wechsler, D. (1997). *Wechsler Adult Intelligence Scale–Third Edition: Administration and scoring manual*. San Antonio, TX: The Psychological Corporation.

Wolff, P. H., Gardner, J., Paccia, J., & Lappen, J. (1989). The greeting behavior of fragile X males. *American Journal on Mental Retardation, 93,* 406–411.