Prelinguistic Communication and the Acquisition of Verbal Communication in Young Children with Fragile X Syndrome

BY

Jennifer L. Flenthrope

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Nancy C. Brady, PhD

Chair

Committee members* ____________________________

Debora B. Daniels, PhD*

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Kathy S. Thiemann, PhD*

Date defended:_______

The Thesis Committee for Jennifer L. Flenthrope certifies that this is the approved version of the following thesis:

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Abstract

This retrospective study investigated potential predictors of the acquisition of verbal communication skills in young children with fragile x syndrome (FXS). In addition, descriptive information concerning this population's early communication development was gathered. The present study was part of a larger FXS research project conducted by researchers from the University of Kansas and the University of North Carolina Chapel Hill. Potential predictors examined included different types of gestures, different communicative functions, characteristics of autism, and overall rates of communication. Predictor data were derived from standardized test scores and video sample analysis. Outcomes examined included standardized test scores, mean length of utterance, and number of different words used during a second observation period. No significant correlations were found between predictor and outcome variables for the whole sample; however, some noteworthy correlations between predictors and outcomes were found for females and participants with high autism symptomatology. Valuable descriptive information was also obtained.

Chapter 1: Introduction

Fragile x syndrome (FXS), which is caused by the mutation of a gene on the X chromosome, is the most common known cause of inherited intellectual disability. The level of disability resulting from FXS ranges from learning disabilities to severe mental retardation. FXS is also the most common known cause of autism. Delays in speech and language development are common symptoms of FXS (National Fragile X Foundation, 2006).

Language Development among Young Children with Fragile X Syndrome

Research on the language development of children with FXS is fairly limited and, historically, has focused primarily on school-age children. The majority of children with FXS are still in the early stages of language development by the time they turn three, and some may not be talking at all yet. Roberts, Hatton, and Bailey (2001) reported that a sample of 26 young boys with FXS used their first spoken words at 28 months of age on average with a range of 9 to 88 months, indicating a severe delay in the onset of spoken language among many children with FXS. Roberts, Mirrett, and Burchinal (2001) found that, in children with FXS, receptive language developed more quickly than expressive language, which developed at about one third of the rate of typically developing children. Thus, expressive language is an area of particular concern for children with FXS, yet not much research has examined early stages of expressive language development in children with FXS. Descriptive information regarding prelinguistic and early verbal communication among this population would be useful in guiding clinicians in the formulation and implementation of treatment plans with these children.

Fragile X Syndrome and Autism

Because males only have one X chromosome, they are usually more affected by FXS than females (Hagerman, 2007, p. 28). Autism occurs in about 25-33% of males and in about 5-15% of females with FXS (Hagerman, 2007, p. 34). Philofsky, Hepburn, Hayes, Hagerman, & Rogers (2004) found that children with FXS who also have autism have more severe receptive and expressive language impairments than children who only have FXS. Descriptive information regarding the characteristics of the prelinguistic and early verbal communication of children with a concurrent diagnosis of FXS and autism would be useful in guiding interventionists planning a treatment program for such children.

Influence of Autism

Autism is a neurodevelopmental disorder characterized by impairments in social interaction, impairments in communication, and repetitive and stereotyped behaviors (APA, 2000). From very early in their development, children with autism show differences in their intentional communication. Babies who are later diagnosed with autism have been found to demonstrate two major differences from typically developing babies. These differences include a lack of joint attention behavior and an abnormal response to human faces and voices (Chawarska and Volkmar, 2005). These young children with autism do not communicate to share focus as typically developing children do; however, they do communicate to express wants and needs (Wetherby, Woods, Allen, Cleary, Dickinson, & Lord, 2004). Stone, Ousley, Yoder, Hogan and Hepburn (1997) found that, when compared to developmentally delayed same age peers with comparable mental ages and expressive vocabularies, two and three-year-old children with autism demonstrated a decreased likelihood to point, show objects, or use eye gaze to communicate. Children with autism in this study also used less complex combinations of behaviors to communicate, and they were observed to request more often and comment less often that their matched peers. In place of developing conventional means of communication, children with autism may develop maladaptive, unconventional or idiosyncratic behaviors to communicate (Wetherby et al., 2004). The presence and severity of autism symptoms varies among children with FXS. Due to the nature of autism and the effects it has on language development, it is necessary to take into consideration the variance in the presence and severity of symptoms of autism among children with FXS when studying this population's language development. Children with FXS who have more symptoms or more severe symptoms of autism may develop language differently from children with FXS who do not have autism. For example, there may be differences in the forms or functions of these subgroups' communication in addition to the known differences in severity of impairment.

Gesture use in typically developing children

By about 12 months of age, gestures become a primary means of communication for the typically developing child. Gestures including giving, showing, and reaching appear when children are about 8 months old, and pointing appears when children are about 12 months (Brady, Bredin-Oja & Warren, 2007, p. 174). In typically developing children, pointing at distal locations has been shown to predict subsequent language development (Brooks & Meltzoff, 2008, p. 217). That is, children who pointed during an assessment completed when children were 11 months old had significantly faster vocabulary development through two years of age than did children who did not point during the assessment. Butterworth and Morisette (1996) found that age of pointing onset predicts gesture use and sound comprehension at 14.4 months. Specifically, children who began pointing at a younger age produced more gestures and understood more animal sounds at age 14.4 months than children who began pointing later, which indicates that pointing may be an important predictor of receptive and expressive communication development. According to Brady et al. (2007), pointing appears to be crucial in the transition between communicating about the immediate environment and communicating about distal events, as mature forms of pointing allow a child to communicate about distant objects. Therefore, children with expressive language delays may benefit from intervention specifically targeting gesture development, particularly the development of pointing.

Types of Gestures

Common gestures can be classified into various categories depending on their forms. According to Bates (as cited in Iverson & Thal, 1998, p. 60), deictic gestures are those that establish reference by calling attention to or indicating an object or event. Deictic gestures can be divided into contact and distal gestures. Leading an adult by the hand or touching an object while pointing at it are examples of contact gestures while pointing at an object from a distance or holding up an object to show it to someone are examples of distal gestures. Representational gestures, according to Crais, Day Douglas and Campbell (2004), establish reference and indicate a particular semantic content. Representational gestures can be divided into object-related and conventional gestures. Object-related gestures are semi-iconic, as this type of gesture indicates some specific feature of the object indicated (Acredolo & Goodwyn, 1988, p. 452). Pantomime is considered an object-related gesture. Conventional gestures represent an action or concept rather than a specific concept (Crais, Day Douglas, & Cox Campbell, 2004). Waving and shrugging one's shoulders are both examples of conventional gestures.

Gesture use in children with Fragile X Syndrome

Although little research has been devoted to prelinguistic communication in children with FXS, one study by Roberts, Mirrett, Anderson, Burchinal and Neebe (2002) found that a sample of 22 males with FXS between 21 and 77 months with a developmental age younger than 28 months demonstrated a significant delay in both conventional gestures and distal gestures. Considering the high rate of autism in children with FXS, findings in relation to gesture use in children with autism should also be considered. According to Brady et al. (2007), disturbances in gesture development is a primary prelinguistic characteristic of children with autism. Children with autism have been found to have large deficits in their use of conventional gestures when compared to typically developing children and moderate deficits compared to developmentally delayed children (Wetherby, Watt, Morgan & Shumway, 2007). In addition, autism has been found to be associated with a decreased *variety* of social interaction gestures used in 9-12 month old infants (Colgan et al., 2006). Additional information regarding how gesture use develops and transitions into verbal language among the FXS population could aid interventionists in creating treatment programs targeting prelinguistic communication. Based on Colgan et al.'s findings, further knowledge of the variety of gestures used by children with FXS would be of value as well.

Communication Functions

Prelinguistic and verbal communicative acts can be classified based upon the act's function. Bruner (1981) identified three broad categories of communication functions that typically develop in a child's first year of life. These include behavior regulation, which is communicating to regulate another's behavior; joint attention, which is communicating to direct another's attention to an object or event; and social interaction, which is communicating to gain or maintain another's attention to oneself. Previous research has shown that rate of communication for joint attention and rate of communication for social interaction each predict expressive and receptive language development in typically developing children (Mundy, Kasari, Sigman, & Ruskin, 1995; Mundy, Sigman, Kasari, & Yirmiya, 1988).

Prelinguistic Predictors of Language Growth in Children with Developmental Disabilities

Previous research has identified a number of prelinguistic factors that appear to predict language growth in children with disabilities. Smith, Mirenda, and Zaidman-Zait (2007), in a study of 35 children with autism between 20 and 71 months whose initial expressive vocabularies were less than 60 words, found that the number of gestures to initiate joint attention used by a child upon entrance into the study was associated with rapid expressive vocabulary growth over a two year time period. Rate of prelinguistic communication has also been demonstrated to b e a good predictor of language growth in children with developmental disabilities. Calandrella and Wilcox (2000) found that rate of prelinguistic communication in a group of developmentally delayed children between 17 and 38 months was significantly correlated with rate of symbol use, rate of different symbol use, and receptive and expressive language scores on a standardized assessment 12 months later. Certain types of gestures have also been demonstrated to be good predictors of language development in children with developmental disabilities. Brady, Marquis, Fleming and McLean (2004) found that, in a group of 18 children with developmental disabilities and vocabularies of ten or fewer words, the children's level of gestural attainment, rate of communication, and parent response contingency were significant predictors of language development over a two year period. Specifically, Brady and colleagues found that the presence of pointing during a relatively brief assessment predicted rate of change in the child's expressive language, as well as the child's initial level of expressive language. None of these studies focused particularly on children with FXS.

The Present Study

In light of a developmental perspective and a continuous view of development, the present study investigated potential prelinguistic predictors of language growth specifically in children with FXS who were non-verbal at about age two years, one month. Some of these children also had autism diagnoses. Unlike other studies examining early communication in children with FXS, females with FXS were also included. The present study investigated the role of several communication skills, including gesture use, overall rate of non-imitative communication, and pragmatic function, in predicting the successful development of verbal communication skills approximately two and a half years later. These communication skills were examined in the context of natural interactions. Standardized test scores as well as data obtained through the analysis of parent-child interactions were examined as outcome variables.

Hypotheses

The following hypotheses were posed:

Hypothesis 1: that higher rates of intentional communication at time one would be associated with higher measures of expressive language at time two.

Hypothesis 2: that a greater variety of gestures at time one would be associated with higher measures of expressive language at time two.

Hypothesis 3: that the presence of pointing at time one would be associated with more developed expressive language skills at time two.

Hypothesis 4: that a higher frequency of communication for joint attention purposes would be associated with more developed expressive language skills at time two.

Hypothesis 5: that high autism symptomatology would be associated with lower scores on all measures of expressive language at time two.

In addition, the present study sought to describe the prelinguistic communication and verbal language acquisition of very young children with FXS. Because the majority of research on children with FXS has focused on school-aged children, there is little descriptive information available about this population's communication when they are very young. Therefore, the acquisition of descriptive information about this population's communication development was also a primary purpose of the present study.

Chapter 2: Methods

Subjects

Participants were young, nonverbal children (24 males and 7 females) with FXS living in the United States. Upon entering the study, these children were observed to use five or fewer different words. On average, participants were approximately two years, one month old during the first observation period with an age range of 11 to 48 months and approximately four years, nine months during the second observation period with an age range of 40 to 76 months. Although formal autism diagnoses were not recorded for the various participants, scores from The Childhood Autism Rating Scale (CARS) obtained at period one indicated that 10 of the participants would qualify for a diagnosis of autism, and CARS scores obtained at period two indicated that 19 of the participants would qualify for a diagnosis of autism. For the purpose of creating subgroups, an average of CARS scores was used with a score of 30 or above indicating placement in the high autism symptomatology subgroup. Three out of seven female participants were placed in the high autism symptomatology subgroup while 15 out of 24 male participants were placed in the high autism symptomatology subgroup. Participants came from a variety of social, racial, and ethnic backgrounds. Recruitment efforts included advertising at national conventions, using a national research registry, networking with FXS family support groups, and advertising via a FXS parent list serve. Please refer to table 1 for additional participant information. Mean composite scores and ranges for the Mullen Scales of Early Learning, the Childhood Autism Rating Scale, and the Vineland Adaptive Behavior Scale are included. The Mullen is an assessment of early cognitive and motor development composed of five subscales, including gross motor, fine motor, visual reception, receptive language and expressive language. The Childhood Autism

Rating Scale is a behavior rating scale that measures autistic behavior in young children, and the Vineland is a measure of personal and social skills, including the domains of communication, daily living skills, socialization, and motor skills.

Table 1

Summary of Participants' Standardized Test Scores and Ages for Time One and Time Two

Observation period	Chronological age in months(mean and standard deviation)	Mullen composite score (mean and standard deviation)	CARS (mean total score and range)	Vineland, Adaptive Behavior Score (mean and standard deviation)
Time 1	Mean=25.23 SD=9.54	Mean=53.81 SD=10.77	Mean=26.97 range=16-42	Mean=68.58 SD=13.06
Time 2	Mean=56.94 SD=8.88	Mean=52.10 SD=9.62	Mean=30.15 range=18-40	Mean=55.48 SD=14.07

Data collection

Larger FXS Study

In the context of the larger FXS study conducted by researchers at the University of Kansas and the University of North Carolina Chapel Hill, data were gathered when children entered the study at an average age of two years, six months, as well as at 18 month intervals until the children were approximately five years, six months. Data included the results of several standardized measures described above, including the Mullen Scales of Early Learning, the Vineland Adaptive Behavior Scale, and the Childhood Autism Rating Scale. In addition, video recordings were made of each child interacting with his or her mother in both structured and unstructured settings, including book reading, making and eating a snack, free play, and activities of daily living.

Present Study

The present study was primarily concerned with the first and last data points from the larger study, which will be referred to as observation period one and observation period two in reference to the present study. In regard to observation period one, scores from The Childhood Autism Rating Scale and data obtained from the analysis of video recordings were examined. Five minute samples from each of the aforementioned parent-child activities (i.e., book reading, making and eating a snack, free play, and activities of daily living) were analyzed to determine each child's overall rate of gesture use, the different functions of each child's communicative acts, the different types of gestures the child used, and the child's total rate of intentional communication. Two samples of activities of daily living were analyzed; thus, 25 minutes of video were analyzed for each participant. All child communication, including gesture use in general, and the functions of each child's communicative acts had been coded as part of the larger FXS study. All communicative acts were designated as serving one of the following functions: social interaction, joint attention, or behavior regulation.

Gesture type coding. Additional coding of gesture types was completed as part of the present study. Gesture types examined included both deictic and representational gestures, including the following: pointing from a distance, pointing while touching the indicated object, touching an adult for a specific communicative function (e.g., leading by the hand), reaching for an object, pushing away an object, giving an object to an adult, showing an object to an adult, tapping an object to attract an adult's notice, moving objects away from or towards an adult, pantomime, and conventional gestures, including waving, shrugging shoulders, shaking head, etc. (see table 2 for brief definitions).

Table 2

Descriptions of Various Gesture Types Coded in the Present Study

Type of Gesture	**Gesture Description**
Distal Point	The index finger is extended towards the object/person of interest, and is 6" or more from the object. Other fingers are curled under.
Touching the Adult	Touching an adult with clear communicative intent.
Showing an Object	Child extends the object toward the adult with the sole intention of showing the object.
Giving an Object	Child gives an object to adult. Child must release the object, and the adult must accept the object.
Tapping	Child taps the object while sharing attention with the adult. Tapping may be with whole hand or with a finger.
Contact Point	Child touches an object with the index finger. *At least two of the adjacent fingers should be curled under or arched up.*
Abbreviated Reach	Child reaches for an object but does not directly grab it. There should be *a momentary, expectant pause by the child*.
Moving Object Away	The child *distinctly and purposefully* moves the object away from the adult.
Pantomime	Pantomime is the use of a part of the body or face to imitate an object or the use of an object.
Raising Arms Up	Child raises his or her arms up to indicate that he or she would like to be picked up.

Moving Object Toward	The child *discreetly and purposefully* moves an object toward the adult.
Throwing/Dropping	This act must have meaning in order to be coded as a gesture. Back and forth ball games should not be coded as gestures.
Waving	Child waves the hand or arm, can be directed towards an object or a person (for the purpose of greeting or saying goodbye).
Shrugging Shoulders	A shrug includes lifting of the shoulders to the ears or upturning of palms to indicate "what" or "I don't know."
Upturned Palm	The palm should be upturned as if to say "give that to me."
Both Palms Upturned	Both hands are held palms up and to the side as if to say/ask "Where is ___?"
Come Here	Upturned palm with one or more fingers or the whole hand wiggling to convey the message to come.
Shh	The gesture must be distinctive with the finger held in close approximation to the mouth.
Head Nod/Shake	A head nod or shake must be intended to convey the message "yes" or "no."
Crossed Arms	Child crosses arms over chest to show dissatisfaction—not just a resting stance.
Clapping	Child brings hands together quickly in midline, palms usually touch.
Hands Over Mouth	Child places one or both hands over mouth as if to express surprise.
Patting Chair	Child pats chair, as in "sit by me".
Pushing Object Away	The purpose of this gesture is always rejection or a signal that the child is finished with the object.
Not Otherwise Specified	Gestures that are not on the list but appear to be clearly communicative.

Coding Reliability

Thirty-two percent of all video recordings were coded separately by a second coder to assess inter-reliability. Training of the secondary coder consisted of reading a coding manual (see the Appendix) and practicing coding the gestures of children from the larger FXS study who did not qualify for the present study due to the child having too many words. The secondary coder was considered a reliable coder after having independently coded 3 of the practice videos (25 minutes each) with at least 85% agreement with the primary coder. An agreement was scored when both coders described a gesture with the exact same code, listed in table 2. A disagreement was scored whenever the two coders used different codes to describe a gesture. Percent agreement for each participant was calculated by dividing the number of gestures agreed upon by the total number of gestures used by the participant.

For the 32% of participants whose videos were coded by a secondary coder, the mean percent agreement was 91% with a range of 75% to 100%. The mean for overall percent agreement was calculated by summing participants' percents of agreement and dividing by the number of participants coded by both coders.

Observation Two Measures

Observation period two communication variables analyzed included expressive language scores from the Mullen, number of different words used, total rate of communication, and mean length of utterance (MLU). The latter three pieces of data were obtained from examining the previously described 25 minutes of videotaped parent-child interaction from time 2. These data were collected by other researchers also working on the University of Kansas FXS research project. Reliability for the number of different words, MLU, and total rate of communication were estimated by this team of researchers using the intraclass correlation coefficient (ICC), (Shrout & Fleiss, 1979), which assesses the inter-rater effect on participant's scores in relation to the individual subjects' effect on scores. The ICCs for rate of number of different words were .987 at time one and .993 at time two. The ICCs for total rate of communication were .894 at time one and .960 at time two, and the ICC for MLU at time two was .983.

Behavioral Coding

The Observer XT by Noldus was used in the coding of parent-child interaction videos, as well as in the analysis of all video recordings. Gestures in general had been coded previously as part of the larger FXS study for all parent-child interaction video recordings using The Observer. Coders for the present study located each of these previously coded gestures and determined specifically what type of gesture was performed. The original FXS study's Observer configurations were created on a previous version of the program (Observer 5.0); however, this version of the program did not allow for the type of coding necessary for the present study. Therefore, the original FXS study's configurations were transferred to the Observer XT where they were updated to include codes for the aforementioned gesture types. In the Observer XT, coders were able to open previously coded video files and their accompanying data files. Coders were then able to view the video files and add the additional codes for gesture types. As gestures in general had already been coded by researchers in the larger FXS study, coders in the present study simply located these previously coded gestures and determined the type of gesture that was used. Please refer to figure 1 for an example of how the Observer appeared on computers used in video sample analysis. Observations to be recoded were selected from the window on the left. A video window appeared in the upper-right corner of the screen (not pictured in figure). Playback controls for reviewing the video file were positioned below the video window. By selecting a previously coded act from the window in the upper-center of the screen, the video file advanced to the time frame in which the act occurred. After reviewing the act, coders selected the appropriate gesture type from the coding configurations window, which is found in the lower-center of the screen.

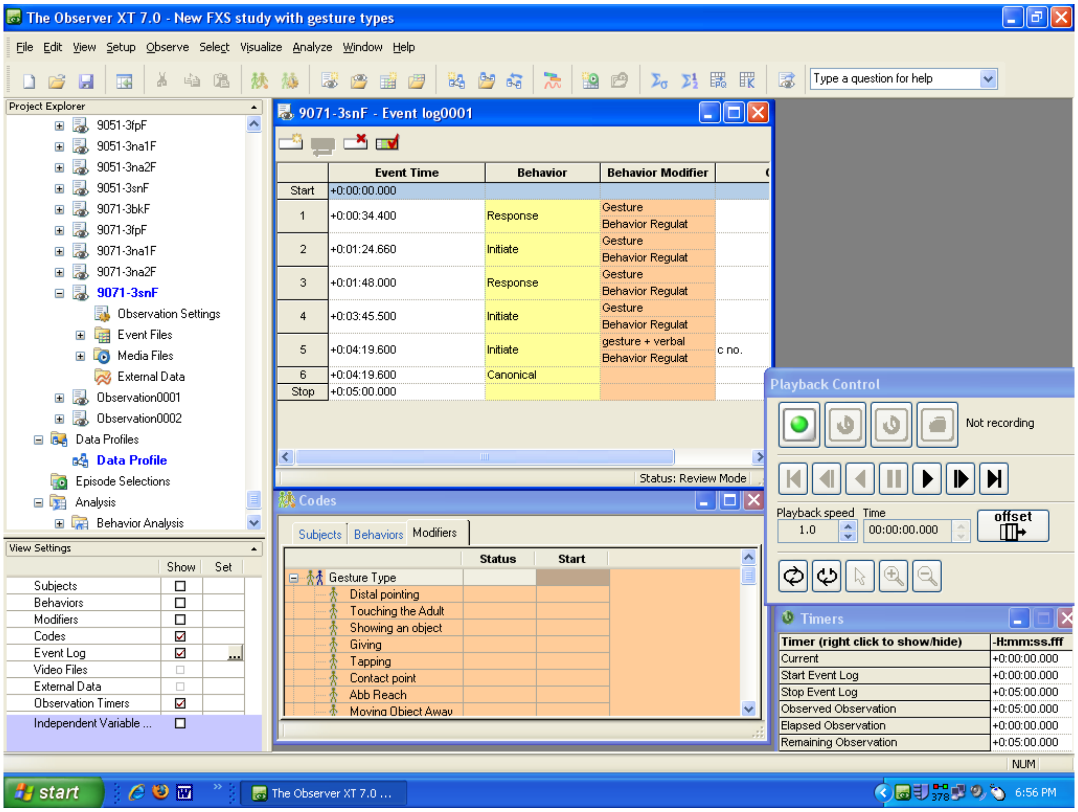

Figure 1. Observer XT behavior coding screen without participant video window (normally in upper right corner).

Behaviors coded specifically for the present study included both deictic and representational gestures. Contact gestures coded included touching an adult for a specific communicative function (e.g., leading by the hand), pointing while touching the object indicated, tapping an indicated object, pushing away an offered object, throwing an object with communicative intent, and giving an object to an adult. Distal gestures coded included pointing from a distance, reaching for an object, moving an object toward or away from an adult, raising arms up to be picked up, and showing an object to an adult. Pantomime was coded as an object-related gesture. Conventional gestures coded included waving, shrugging shoulders, making a "shh" gesture, shaking/nodding head, expectantly displaying an upturned palm (as if stating, "give it to me"), displaying two upturned palms (as if asking "where"), signaling come here by wiggling one or more fingers of an upturned palm, clapping, covering mouth as if surprised, crossing arms to show resistance, and patting a chair (as if requesting "sit by me"). Gestures (see table 2) were only coded as communicative if they were accompanied by attention to the adult and showed joint reference. Attention to the adult was demonstrated in a number of ways, including intentionally touching the adult, looking at the adult, showing an object to the adult, giving an object to the adult, or moving an object toward the adult. In specific cases in which there was clear joint reference between the child and mother, such as shared book reading, attention to the adult was implied.

Chapter 3: Results

Correlation Data for the Whole Group

Table 3 displays correlations between predictor and outcome variables. Predictor variables included number of different gestures, rate of communication for joint attention, and CARS total scores for time one. Outcome variables included MLU, Mullen expressive language raw scores, and rate of different words used for time two. Additional variables were included in table 3 to provide further information regarding time one and time two relationships. In table 3 and all subsequent correlation tables, outcome measures are highlighted, and outcome measure correlations are shaded. As mentioned previously, no significant correlations were found between predictor and outcome variables for the whole sample.

Other correlations (denoted by asterisks) in table 3 were significant; however, these significant correlations were generally between related variables (e.g., total rate of communication and rate of different words used) for the same time period. For example, rate of total communication at time one was found to be significantly correlated with number of different gestures, Mullen expressive language raw scores, and rate of different words all at time one. This indicates that children who communicated more with all forms of communication used a wider variety of gestures and a higher rate of different words. These children also performed better on the expressive language subtest of the Mullen. Similarly, a significant positive correlation was found between rate of different words and Mullen expressive language raw scores at time one, which indicates that children who performed better on the Mullen expressive language subtest also used a higher rate of different words. As expected, CARS totals for time one and time two were also significantly positively related, indicating some consistency in the severity of autism symptomatology in participants over time. Significant relationships were not found between time one and time two measures of expressive language (i.e., Mullen expressive language raw score, rate of different words, and rate of total child communication), indicating that children who demonstrated relatively strong expressive language skills at time one did not necessarily demonstrate strong expressive language skills at time two. Significant positive correlations were found between nearly all time two variables with the exception of CARS totals and rate of total communication and CARS totals and expressive language raw scores from the Mullen. These two correlations were significant but negative, indicating that participants with higher autism symptomatology communicated less frequently and performed more poorly on the Mullen expressive language subtest. The positive correlations between other time two variables indicate that the various measures of expressive language are strongly related, which is to be expected since they are measures of the same construct.

Table 3

Pearson's Correlations for Time 1 and Time 2 Variables for All Participants

Measure	1	2	3	4	5	6	7	8	9	10	11	12
1. T1 Number of Different Gestures		-.03	.05	.27	.08	.51**	-.12	-.22	.05	-.06	-.11	-.12
2. T1 Rate of Joint Attention			-.10	.02	-.02	-.09	-.18	.28	.20	-.13	-.08	.04
3. T1 Mullen Expressive Language Raw Score				.11	.50**	.58**	-.24	-.25	.01	-.17	-.16	-.05
4. T1 CARS Total					.41	.32	-.28	-.28	.57**	-.24	-.23	-.31
5. T1 Rate of Different Words						.55**	.05	.01	-.10	.09	.19	.12
6. T1 Rate of Total Child Communication							-.19	-.31	.05	-.15	-.15	-.04
7. T2 Rate of Total Child Communication								.85**	-.63**	.89**	.90**	.74**
8. T2 Rate of Joint Attention									.51**	.82**	.85**	.75**
9. T2 CARS Total										-.68**	.62**	.62**
10. Mullen Expressive Raw Score											.94**	.84**
11. T2 Rate of Different Words												.86**
12. T2 Mean Length of Utterance												

*. Correlation is significant at the 0.05 level (2-tailed).

**. Correlation is significant at the 0.01 level (2-tailed).

Weak to moderate relationships between participants' time one CARS scores and expressive language outcomes were found with the strongest correlation (r= -.31) between time one CARS scores and time two MLU. Please refer to figure 2 for a graphical depiction of this relationship.

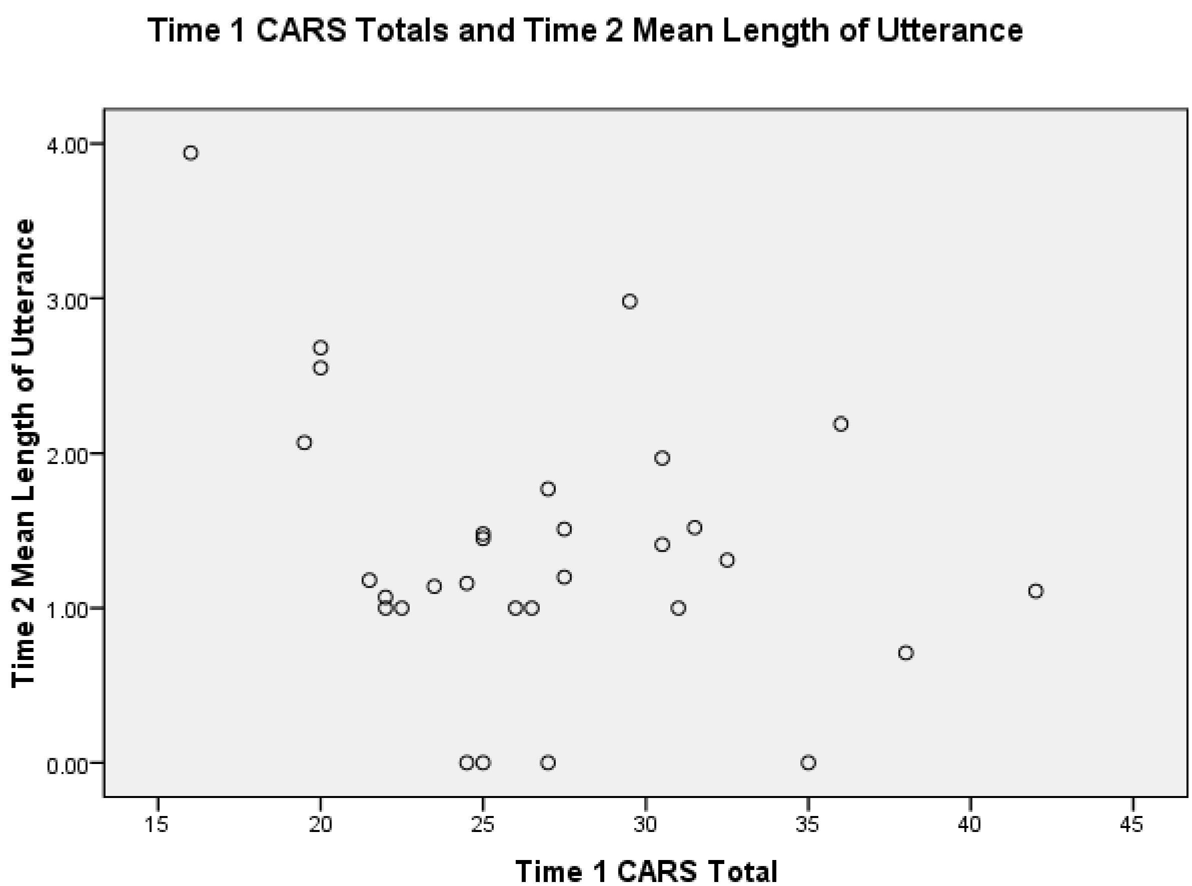

Figure 2. Scatter plot depicting the relationship between time 1 CARS total scores and time 2 mean length of utterance among all participants.

Similar yet slightly weaker relationships were found between time one CARS total scores and number of different words used (r= -.23) and Mullen expressive language raw scores (r= -.24).

Multiple regression analysis was intended to be used to determine if children's overall rate of intentional communication, use of a variety of gestures, use of pointing, rate of communication for joint attention, and autism diagnosis predict expressive language development, as determined by MLU, number of different words used, and expressive language scores on the Mullen. However, in anticipation of these analyses, Pearson's correlations were performed between the predictor and outcome variables. No significant correlations were found between predictor and outcome variables for the whole sample. Therefore, multiple regression analyses did not appear warranted. Exploratory multiple regression analyses indicated that none of the predictor variables accounted for significant amounts of additional variance, above the variances accounted for by developmental and chronological ages. Therefore, we decided to focus on the correlational data in order to consider the relationships between various predictor and outcome variables.

Although no significant correlations were found between predictor and outcome variables for the whole sample, numerous significant correlations existed between chronological age and communication measures at time one and time two. Due to the quantity of significant correlations existing between time one and time two variables and chronological age, these correlations were placed in separate tables below (see tables 4 and 5).

Table 4

Pearson's Correlations for All Participants for Chronological Age at Time 1

Measure	Age at T1
T1 Number of Different Gestures	.45*
T1 Rate of Joint Attention	-.01
T1 CARS Total	.56**
T1 Mullen Composite	-.66**
T1 Mullen Expressive Language Raw Score	.63**
T1 Rate of Different Words	.41*
T1 Rate of Total Child Communication	.55**

*. Correlation is significant at the 0.05 level (2-tailed).

**. Correlation is significant at the 0.01 level (2-tailed).

Table 5

Pearson's Correlations for All Participants for Chronological Age at Time 2

Measure	Age at T2
T2 Rate of Joint Attention	-.32
T2 CARS Total	.43*
T2 Mullen Composite	-.42*
T2 Mullen Expressive Language Raw Score	-.31
T2 Mean Length of Utterance	-.33
T2 Rate of Different Words	-.23
T2 Rate of Total Child Communication	-.41*

*. Correlation is significant at the 0.05 level (2-tailed).

**. Correlation is significant at the 0.01 level (2-tailed).

At time one, significant positive correlations were found between age and all expressive communication measures, including number of different gestures, Mullen expressive language raw score, rate of different words, and rate of total communication. Yet, at time two, age is negatively correlated with all of the communication measures, including mean length of utterance, rate of joint attention, Mullen expressive language raw score, rate of different words, and rate of total child communication. These negative correlations appear to be due to the fact that several of the older participants' communication measures did not change much between time one and time two. For both time one and time two, negative significant correlations were found between Mullen composite scores and age. In regard to time one, in light of the strong positive correlation between age and Mullen expressive language raw scores, the negative correlation between age and Mullen composite scores appears to be attributable to older children scoring lower on other subtests, such as fine motor, visual reception and/or receptive language. In regard to time two, the significant negative correlation between age and Mullen composite scores again appears to be due to several older children's scores not changing much between time one and time two. At both observation periods, age was found to be significantly positively correlated with CARS totals, which means that older participants had more symptoms of autism.

Correlation Data for the High and Low Autism Symptomatology Subgroups

Because of the special challenges to language learning presented by symptoms of autism, participants were divided into two subgroups based upon an average of their CARS scores with children whose average score was greater than or equal to 30 (the clinical cut-off for an autism diagnosis) falling into the high autism symptomatology subgroup. This division into subgroups allows for an analysis of language development taking into consideration the special challenges faced by children with FXS who also have many or more severe symptoms of autism. Of the 24 male participants, 15 were placed in the high symptomatology subgroup, and, of the seven female participants, three were placed in the high symptomatology subgroup. Please refer to tables 6 and 7 for predictor and outcome variables' correlations for the high and low autism symptomatology subgroups.

Table 6

Pearson's Correlations for the Low Autism Symptomatology Subgroup (CARS Scores <30)

Measure	1	2	3	4	5	6	7	8	9	10	11	12
1. T1 Number of Different Gestures		.80**	.59*	.18	.30	.66*	-.11	-.18	.42	-.11	.03	-.09
2. T1 Rate of Joint Attention			.51	.07	.43	.73**	-.20	.21	.13	-.03	.04	.03
3. T1 Mullen Expressive Language Raw Score				-.10	.24	.60*	-.46	-.40	.33	-.22	-.24	-.20
4. T1 CARS Total					.61*	.29	-.22	-.29	.41	-.34	-.19	-.44
5. T1 Rate of Different Words						.74**	-.06	-.09	.16	.07	.16	.01
6. T1 Rate of Total Child Communication							-.19	-.27	.32	-.06	-.05	-.08
7. T2 Rate of Total Child Communication								.95**	-.51	.81**	.82**	.57*
8. T2 Rate of Joint Attention									.61*	.91**	.91**	.68*
9. T2 CARS Total										.72**	.55*	-.55
10. Mullen Expressive Raw Score											.94**	.81**
11. T2 Rate of Different Words												.84**
12. T2 Mean Length of Utterance												

*. Correlation is significant at the 0.05 level (2-tailed).
**. Correlation is significant at the 0.01 level (2-tailed).

Table 7

Pearson's Correlations for the High Autism Symptomatology Subgroup (CARS scores ≥ 30)

Measure	1	2	3	4	5	6	7	8	9	10	11	12
1. T1 Number of Different Gestures		-.15	-.10	.49*	-.06	.46	-.32	-.41	.24	-.21	-.40	-.37
2. T1 Rate of Joint Attention			-.17	-.01	-.09	-.21	-.22	.51*	.42	-.17	-.10	.11
3. T1 Mullen Expressive Language Raw Score				.11	.62**	.59*	-.16	-.19	-.37	-.12	-.10	.09
4. T1 CARS Total					.52*	.35	.09	.02	.21	.28	.15	.20
5. T1 Rate of Different Words						.47	.09	.03	-.30	.07	.20	.19
6. T1 Rate of Total Child Communication							-.18	-.34	-.23	-.19	-.21	.04
7. T2 Rate of Total Child Communication								.69**	-.36	.90**	.94**	.73**
8. T2 Rate of Joint Attention									-.03	.65**	.72**	.69**
9. T2 CARS Total										-.26	-.35	-.10
10. Mullen Expressive Raw Score											.91**	.73**
11. T2 Rate of Different Words												.77**
12. T2 Mean Length of Utterance												

*. Correlation is significant at the 0.05 level (2-tailed).

**. Correlation is significant at the 0.01 level (2-tailed).

Although there were no significant correlations between predictor and outcome variables among the high and low autism symptomatology subgroups, some interesting relationships were found. For example, a moderate negative relationship was found between the number of different gestures used at time one and the rate of different words used at time two among the high autism symptomatology subgroup. Please refer to figure 3 for a graphical depiction of this relationship.

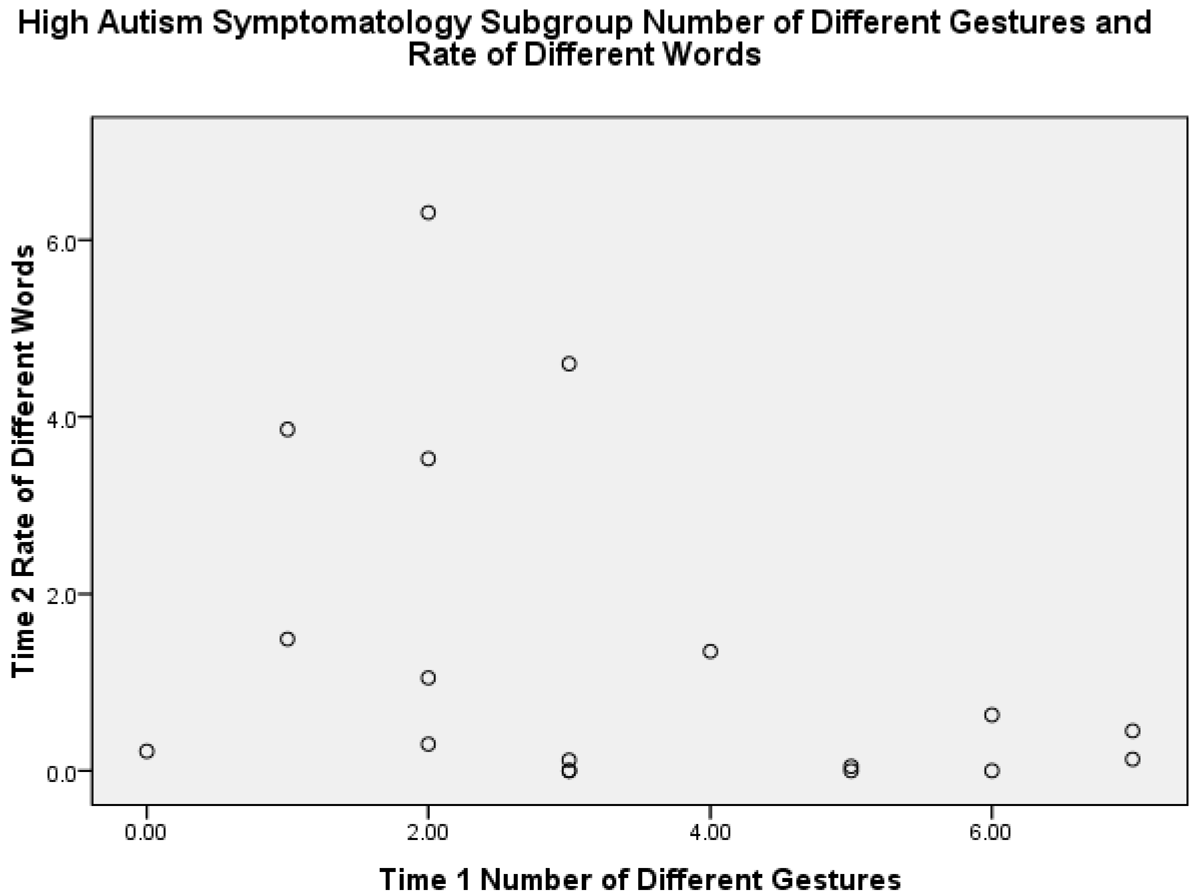

Figure 3. Scatter plot depicting the relationship between the number of different gestures used at time 1 and the rate of different words used at time 2 among participants in the high autism symptomatology subgroup.

Correlation Data for the Male and Female Subgroups

Because the majority of research on children with FXS has focused on male children, the present study divided males and females into two separate subgroups so that any gender-dependent differences might be identified. Information regarding communication among female children with FXS is of great value simply because there is very little information in the literature about this subgroup. There were seven female participants and 24 male participants. Please refer to tables 8 and 9 for predictor and outcome variables' correlations for the male and female subgroups.

Table 8

Pearson's Correlations for Predictor and Outcome Variable for Male Subgroup

Measure	1	2	3	4	5	6	7	8	9	10	11	12
1. T1 Number of Different Gestures		-.10	-.07	.32	.11	.47*	-.16	-.34	.06	-.08	-.17	-.15
2. T1 Rate of Joint Attention			-.15	-.01	-.06	-.14	-.27	.31	.26	-.21	-.17	0.0
3. T1 Mullen Expressive Language Raw Score				.15	.55**	.54**	-.24	-.27	-.03	-.17	-.15	-.06
4. T1 CARS Total					.39	.37	-.31	-.27	.51*	-.06	-.14	-.09
5. T1 Rate of Different Words						.57**	-.03	-.10	-.21	.05	.14	.10
6. T1 Rate of Total Child Communication							-.19	-.37	-.01	-.16	-.17	-.01
7. T2 Rate of Total Child Communication								.78**	-.64**	.89**	.87**	.66**
8. T2 Rate of Joint Attention									-.39	.74**	.77**	.63**
9. T2 CARS Total										-.57**	-.57**	-.51*
10. Mullen Expressive Raw Score											.93**	.75**
11. T2 Rate of Different Words												.78**
12. T2 Mean Length of Utterance												

*. Correlation is significant at the 0.05 level (2-tailed).
**. Correlation is significant at the 0.01 level (2-tailed).

Table 9

Pearson's Correlations for Predictor and Outcome Variable for Female Subgroup

Measure	1	2	3	4	5	6	7	8	9	10	11	12
1. T1 Number of Different Gestures		.60	.25	-.19	-.04	.58	.21	.28	-.38	.40	.35	.23
2. T1 Rate of Joint Attention			.21	.02	.42	.39	.68	.70	-.32	.67	.81*	.70
3. T1 Mullen Expressive Language Raw Score				-.47	.33	.75	-.10	.01	-.33	.22	.11	.31
4. T1 CARS Total					.53	-.10	-.07	-.13	.54	-.31	-.16	-.45
5. T1 Rate of Different Words						.52	.31	.30	.14	.26	.36	.22
6. T1 Rate of Total Child Communication							-.05	.01	-.03	.17	.14	.12
7. T2 Rate of Total Child Communication								.99**	-.54	.91**	.97**	.87*
8. T2 Rate of Joint Attention									-.66	.96**	.98**	.90**
9. T2 CARS Total										-.79*	-.59	-.69
10. Mullen Expressive Raw Score											.95**	.92**
11. T2 Rate of Different Words												.93**
12. T2 Mean Length of Utterance												

*. Correlation is significant at the 0.05 level (2-tailed).

**. Correlation is significant at the 0.01 level (2-tailed).

As mentioned previously, some noteworthy relationships were found when participants were subdivided by certain characteristics. Specifically, females' rate of joint attention at time one was found to be significantly related to their rate of different words used at time 2. Please refer to figure 4 for a graphical depiction of this relationship.

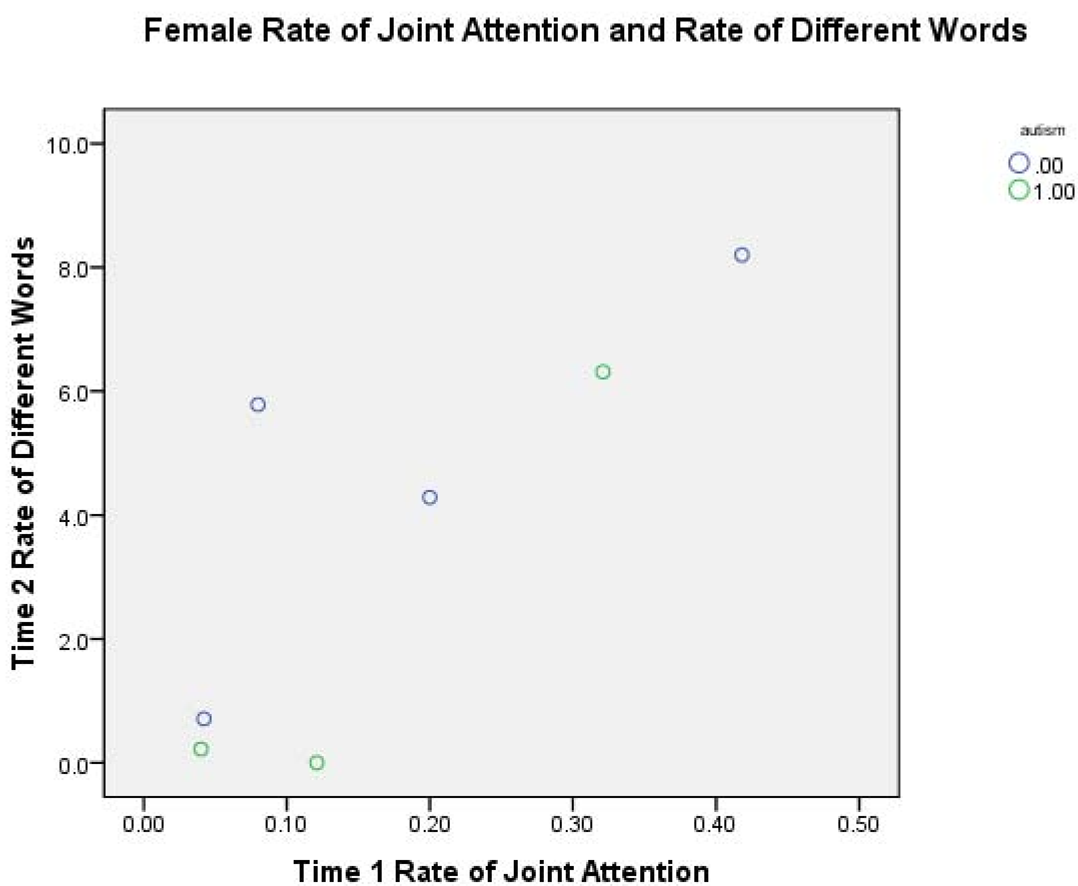

Figure 4. Scatter plot depicting the relationship between female participants' rate of joint attention at time 1 and their rate of different words used at time 2.

Males did not show this same relationship between joint attention and rate of different words used.

Although the relationship between rate of joint attention at time one and rate of different words used at time two among females was the only significant relationship found between predictor and outcome variables, other relationships were strong enough to be of interest. For example, among females, contrary to findings in the high autism symptomatology subgroup, a moderate *positive* relationship was found between the number of different gestures used at time one and the rate of different words used at time two. Please refer to figure 5 for a graphical representation of this relationship.

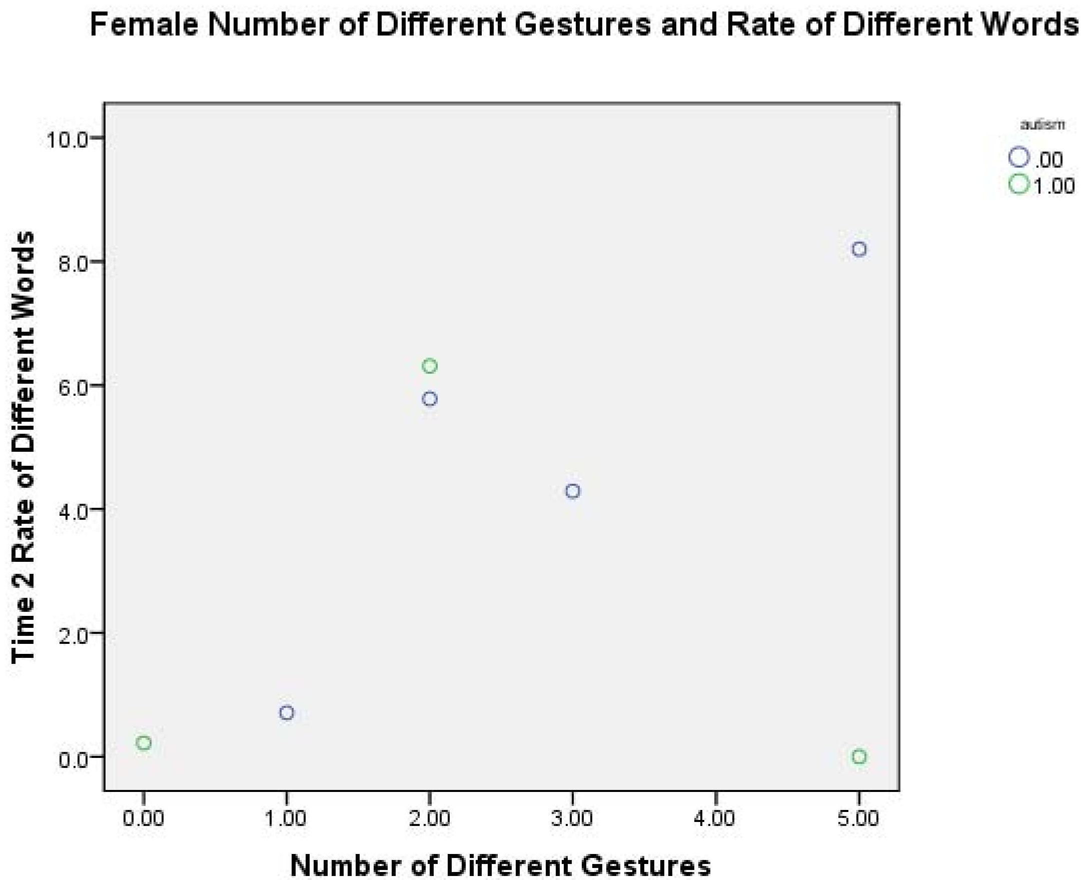

Figure 5. Scatter plot depicting the relationship between female participants' number of different gestures used at time 1 and their rate of different words used at time 2.

Descriptive Results

Descriptive information from both time periods for all participants is also valuable. Information regarding the form and function of participants' communication provides insight into how their communication develops over time.

Communication at Time One

On average, children were two years, one month at time one with an age range of 11 to 48 months. Great variability in communication was observed among participants.

Communication forms. In general, participants were found to communicate primarily using gestures at time period one with an average of 62.5% of their communication consisting of gestures. The proportion of participants' communication consisting of gestures ranged from 0-100%, demonstrating that there was great variability among participants' gesture use; however, of the 31 participants, 21 communicated using gestures at least 50% of the time. An average of 8.3% of participants' communication consisted of verbalizations with a range of 0-66%. Other communication modalities observed included vocalizations and signs; however, the majority of most of the participants' communication consisted of gestures at time 1. Types of gestures observed included both deictic and representational gestures. Contact gestures were observed most frequently; however, distal gestures and conventional gestures were also observed. No object-related representational gestures were observed. Please refer to table 10 for information regarding overall frequency of use and the number of participants who used each type of observed gesture.

Table 10

Frequency of Gesture Use at Time 1

Gesture Type	Number of Children Who Performed This Gesture	Overall Frequency (Range)
Touching Adult	23	1-21
Tapping	13	1-3
Not Otherwise Specified	5	1-2
Pushing Object	15	1-6
Showing	3	1-1
Contact point	5	1-3
Distal point	2	1-1
Raising Arms Up	2	1-3
Nodding/Shaking Head	8	1-3
Giving	14	1-8
Throwing/Dropping	6	1-6
Moving Object Away	3	1-2
Clapping	4	1-4
Abbreviated Reach	3	1-2
Moving Object Toward	6	1-5
Shh	1	1-1

There did not appear to be a strong relationship between the use of any one specific type of gesture at time one and positive verbal language outcomes at time two based upon a comparison of the gestures of children who had a high rate of different words used (i.e., above the median) at time two with those of children who had a low rate of different words used. Overall rates of communication varied greatly among participants for period one. An average of 14 acts with a range of 2-41 acts was observed during the 25 minutes of video analyzed.

Communication functions. Participants' communicative acts were designated as serving one of three communicative functions: joint attention, behavior regulation, or social interaction. At time 1, participants were observed to communicate most often for behavior regulation purposes and least often for social interaction purposes. Please see table 11 for additional information regarding rates of communication for various functions.

Table 11

Rate of Communicative Function Use for Time 1 and Time 2

Communicative Function	**Time 1**	**Time 2**
Joint Attention	Mean=0.17 Range=0-1.37	Mean=3.00 Range=0.20-8.08
Behavior Regulation	Mean=0.35 Range=0-3.50	Mean=1.26 Range=0.30-3.43
Social Interaction	Mean=0.03 Range=0-.41	Mean=0.20 Range=0-1.02

Communication at Time Two

On average, participants were four years, nine months at time two with an age range of 40-76 months. For observation period two, there continued to be great variability among participants.

Communication forms. An average of 121 communicative acts with a range of 9-265 acts was observed during the 25 minutes of video analyzed for period two. The majority of participants were primarily verbal communicators by time two with 52% of participants communicating verbally 50% or more of the time. The average proportion of strictly verbal communication for all participants at time two was 47% while the average proportion of strictly gestural communication for all participants was 37%. Not infrequently during time two, participants were noted to combine gestures with verbal communication with an average of 10% of their communication consisting of this combination of modalities. See figure 6 for a visual depiction of modality usage across observation periods.

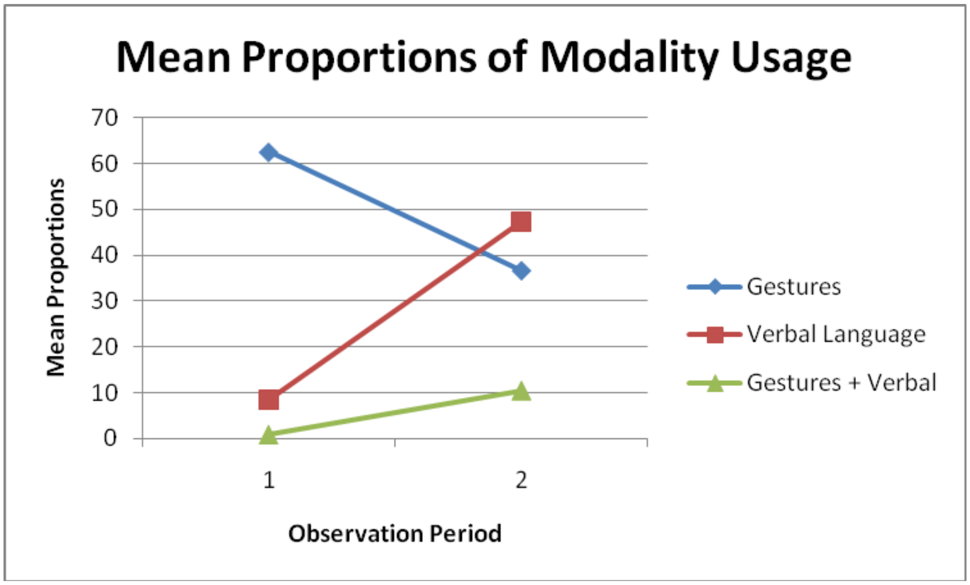

Figure 6. Mean proportions of modality usage across observation periods.

Communication functions. At time two, participants were observed to communicate most often for joint attention purposes and least often for social interaction purposes. Please refer to table 11 above for additional information regarding communication functions at time two.

Chapter 4: Discussion

This study provided heretofore unreported data regarding very early communication produced by young children with FXS. At time one, when these preverbal children were an average of two years, one month old, the majority communicated primarily through gestures; however, by time two, when children averaged four years, nine months old, over half were primarily verbal communicators. At observation one, preverbal children with FXS used a wide variety of gestures, including both contact and distal deictic gestures and conventional representational gestures. No object-related representational gestures were observed. Participants were observed to communicate primarily for behavior regulation purposes at time one and primarily for joint attention purposes at time two. Frequency of communication was also observed to increase greatly from time one to time two for the group as a whole.

Although none of the original hypotheses were supported for the whole sample of participants with a significant correlation, significant support was found for the hypothesis regarding joint attention in the female subgroup. In addition, weaker yet notable relationships between potential predictors and outcomes were found for the group as a whole as well as for some subgroups. The general lack of support for the present hypotheses may be due at least in part to a limited sample size and the variability among participants' ages.

Hypothesis One

In regard to the first hypothesis that higher rates of intentional communication at time one would be associated with higher measures of expressive language at time two, no significant correlations were found. The correlation between rate of total child communication at time one and MLU at time two was -.04 while the correlation with rate of different words used at time two was -.15, and the correlation with expressive language scores on the Mullen at time two was -.15. As noted previously, past research has found that rate of prelinguistic communication is a good predictor of language growth in children with developmental disabilities (Calandrella and Wilcox, 2000), yet the present study found weak negative relationships between rate of prelinguistic communication and later measures of expressive language. These results may be attributable to the variability in participants' ages. At time one, older children were found to communicate at a higher rate; however, at time two, older children were found to communicate at a lower rate and less sophisticatedly (i.e., they had a lower MLU and a lower rate of different words used). At time one, all participants were producing fewer than five words; however, these participants ranged in age from 11 to 48 months. Younger participants may not have been as severely delayed as older participants. Significant negative correlations between chronological age and time two language measures indicate that older participants did not make the same communication gains between time one and time two that younger participants made. Perhaps if all participants were closer in age, the present results would have more closely resembled those of Calandrella and Wilcox and a positive relationship would have been found.

Hypothesis Two

Concerning the second hypothesis that a greater variety of gestures at time one would be associated with higher measures of expressive language at time two, no significant correlations were found for the group as a whole to support this hypothesis. The correlation between a greater variety of gestures and MLU was -.12 while the correlation with rate of different words used was -.11, and the correlation with expressive language scores on the Mullen was -.06. These results suggest that gesture use among toddlers with FXS may not be useful in predicting future communication development, perhaps due to characteristics within the group.

When participants were subdivided by gender, however, a moderately strong yet not significant positive relationship was found between variety of gestures and rate of different words used among females. Contrarily, when participants were subdivided into low and high autism symptomatology subgroups, a slightly stronger though not significant negative relationship was found between these variables. This renders the relationship between variety of gestures used and number of different words used unclear.

Perhaps there are some characteristics unique to each subgroup responsible for their varied results. Children with autism have been found to have large deficits in their use of conventional gestures when compared to typically developing children and moderate deficits compared to developmentally delayed children (Wetherby et al., 2007). In addition, autism has been found to be associated with a decreased *variety* of social interaction gestures used in 9-12 month old infants (Colgan et al., 2006). The difference in results between the female subgroup and the high autism symptomatology subgroup may be due to this decrease in gesture usage among children with autism. Negative correlations among the high autism symptomatology subgroup and the group as a whole may be due in part to the wide age range of participants (e.g., 11-48 months for the whole group; 12-48 months for the high symptomatology group). The female subgroup, however, which showed a moderate positive correlation, was more uniform in age with a range of 11-24 months. Perhaps the positive relationship among variety of gestures and later expressive language outcomes is due to the greater age uniformity in the female subgroup. Differing results may also be due to the fact that female participants were less impaired than male participants who composed most of the high autism symptomatology subgroup. Additionally, it should be noted that older children at time one used a wider variety of gestures than younger children, and older children who qualified for the present study (i.e., had fewer than five words at time one) may have been more severely delayed than younger children. These older children who used a wide variety of gestures may have continued to rely on gestural communication rather than transition to spoken communication.

Hypothesis Three

As regards the third hypothesis that the presence of pointing at time one would be associated with more developed expressive language skills at time two, no analyses were conducted, because only two participants in the study demonstrated distal pointing during the observed time period. One of these participants used a rate of different words above the mean at time two while the other used a rate of different words below the mean at time two. One of these participants only communicated 58 times during the time two observation period while the other communicated 227 times. This would indicate that the presence of pointing during the prelinguistic period does not guarantee rapid verbal language growth; however, it is impossible to make any real judgments based on the extremely limited number of children observed to communicate using distal points. Therefore, the present results in regards to distal pointing cannot justly be compared to previous research on this matter.

Hypothesis Four

In regard to the fourth hypothesis that a higher frequency of communication for joint attention purposes would be associated with more developed expressive language skills at time two, no significant correlations were found to support this hypothesis when all participants scores were included in analysis.

When groups were subdivided by gender, however, a significant correlation (.81) was found for females. No other subdivisions were found to have significant correlations between these variables. When examining these results, several factors should be considered. As mentioned previously, females are generally less affected by FXS than males because they have two X chromosomes. Perhaps the severity of FXS is an overriding factor in determining the development of verbal communication, despite early rates of communication for joint attention purposes. Females were also, on average, much younger than males at time one and time two observations with the average age difference between genders being 11 months at time one and 19 months at time two. At time one, the average age for females was 16.7 months with a range of 11-24 months, and the average age for males was 27.8 months with a range of 12-48 months. Perhaps communication for joint attention is a better predictor of future language development when children are younger, or perhaps the wide range in ages among the male subgroup is partially responsible for the differences in results. Sample size should also be considered. The female subgroup was very small (N=7). Although findings from the female subgroup support previous research concerning joint attention and subsequent language development (Mundy, Sigman, Kasari, & Yirmiya, 1988), the very small number of girls included raises some questions as to the validity of the results.

Previous research (Smith et al., 2007) has found that the number of gestures to initiate joint attention used by young children with autism was associated with rapid expressive vocabulary growth over a two year time period, but, in the present study, a significant relationship between communication for joint attention at time one and measures of expressive language at time two was not found for the autism subgroup. Perhaps that is due in part to the fact that all communication for joint attention purposes was included in this study and not just gestures to initiate joint attention. This difference in results may also be due in part to the fact that participants in the aforementioned study presumably did not have a diagnosis of FXS. Past research has found that there are more severe receptive and expressive language deficits among children with FXS and autism than among children with only autism (Philofsky et al., 2004). These more severe deficits among children with FXS and autism may be responsible for the present results. In addition, classification in the high autism symptomatology subgroup in the present study was based on an average of CARS score ratings; whereas, classification as having autism in the Smith et al. (2007) study required diagnosis by a multidisciplinary team. Because of this stricter requirement, the participants in Smith et al.'s study may have been a more homogeneous group, which may partially account for the differences between past research and the present results.

Hypothesis Five

Concerning the fifth hypothesis that autism symptoms would be associated with lower scores on all measures of expressive language at time two, no significant correlations were found between the predictor and outcome variables. Philofsky et al. (2004) found that children with FXS who also have autism have more severe expressive language impairments than children who only have FXS, but in the present study, no such significant results were found. Again, the way in which children were divided into the high and low autism symptomatology subgroups may have played a role in the differing results between studies. Children with FXS and autism in the Philofsky et al. study were diagnosed using three separate diagnostic systems (DSM-IV, Autism Diagnostic Interview-Revised, and Autism Diagnostic Observation Schedule) as well as clinical judgment by a psychologist with an expertise in autism. In addition, it should be noted that the average CARS score range for all participants in the present study was 17.75-35; therefore, there may not have been much difference between those in the high autism symptomatology subgroup (score of 30 or above) and those in the low autism symptomatology subgroup (score below 30).

Limitations

When examining the results from the present study, several limitations must be taken into consideration. First of all, the sample size, especially in reference to some subgroups, was quite small, thus limiting the power of the results. In addition, the correlation analysis did not allow for the controlling of intellectual differences. Variability among participants' ages also likely impacted results; although, all participants were essentially nonverbal upon entrance into the study. All communication samples for each observation period were obtained on the same day, which may have negatively affected results if participants had particularly good or bad days when they were observed. Another limitation of the present study is that the type of communication intervention provided to participants was not accounted for or described, as data at time 1 only included hours of service and not details about the type of service provided to participants. Participants' outcomes may have been strongly influenced by the type of intervention they received, but this was not accounted for in the present study. Results in regard to the high and low autism symptomatology subgroups are also weakened due to the fact that participants did not receive a formal evaluation from a multidisciplinary team, and only one diagnostic tool was used in the classification of participants into the high and low subgroups. Perhaps significant relationships would have been found had these two groups been formed based on more stringent criteria. These various limitations may be at least partially responsible for differences between the present study and previous research.

Implications

The present results suggest that gesture use and high autism symptomatology among young preverbal children with FXS may not be strongly related to these children's later language development. Overall rate of intentional communication may not be strongly related to later language development among this population, either. Joint Attention may be related to future verbal language development among young nonverbal females with FXS. One possible application of these results may be for early interventionists to focus treatment plans on increasing children with FXS's amount of communication for joint attention, especially among young females with FXS. Another possible application in light of significant negative correlations between age and later language outcomes would be for intervention to begin at very young ages so as to improve children's chances of positive language outcomes later in life. However, further research with greater attention to the aforementioned limitations of the present study should be conducted before any potential applications suggested by this research are put into practice. Future research should also investigate the subset of children who did not successfully acquire verbal language and try to determine what factors impede language development among this population.

Conclusion

As FXS is the most common known cause of inherited intellectual disability and the most common known cause of autism, the study of its effects is highly relevant for healthcare and special education service providers. In light of the increased rate of autism in the general population, it is of ever greater importance that this disorder and its treatment be better understood. Early intervention has been found to be crucial when working with children with autism; therefore, it is necessary that the early development of this population be thoroughly studied in order to pinpoint those treatments which will prove most effective for these children at a young age. Insights obtained while studying children with FXS and autism may generalize to the autistic population as a whole. In regards to speech and language development, which is generally delayed in children with FXS, it is of great value for service providers to have a good understanding of prelinguistic communication and the acquisition of verbal communication among this population so that they may best aid these children in their development and subsequent academic and social success. For example, children may benefit from receiving intervention specifically targeting a type of gesture or a communicative function if those factors are found to be associated with subsequent rapid language development, or it may be better to focus on early word production or other symbol usage, such as PECS, if those factors are more highly associated with subsequent language development. Better intervention provided to young children may lead to higher functioning school age children, adolescents, and adults. Thus, highly effective treatment at a young age may result in a lesser degree of support and services needed later on in the child's life, which has positive social and financial consequences. Further research into the development of children with FXS is certainly warranted, as the results of such research could have far-reaching effects for more than the population being studied.

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Appendix

Gesture Coding Manual

GESTURES: All gestures **MUST** be visible to be coded. All gestures must be paired with joint referent/adult attention. The coder cannot assume a gesture's occurrence from contextual cues. The hand or other body part performing the gesture must be visible to the coder. If the child performs a series of different gestures without a 3 second pause or interruption by an adult communication act, code the last gesture performed.

1. Distal Point (previously coded as "Distal Point", "point + verbal", or "point + vocal")

The index finger is extended towards the object/person of interest. The other fingers should be at least slightly curled under making the point obvious. The child's finger does not touch the object/person and is 6" or more from the object.

The following gestures were previously coded as "gesture", "gesture + verbal", or "gesture + vocal":

2. Touching the adult

Examples include acts such as placing the adult's hand on a jar to request help opening it, hugging adult, pulling on adult, pushing adult away, and physically manipulating the adult's body. NOT CONSIDERED GESTURES: Sitting in the adult's lap, leaning against an adult, or accidentally brushing the adult.

3. Showing an object to the adult

Child extends the object toward the adult with intention of showing the object. The adult is not expected to take or do anything with the object, except to look and perhaps comment on it.

i. The intention of the act must be solely to "show" the object.

ii. The adult is not expected to take or do anything with or to the object except to look and perhaps to comment upon it.

4. Giving

Child gives an object to adult. Child must release object. Child must demonstrate attention to adult and, extend object toward and release object. For a gesture to be considered a give, it must be received by an adult. If the child is physically unable to reach the adult, the adult may meet the child's extended arm to take the offered object. *If the child uses a Picture Exchange Communication System (PECS), code these as "gestures" then put PECS in the comment section.*

5. Tapping

Child taps the object while sharing attention with the adult. Child seems to want the adult to notice the object. If the intent is rejection, do not code as tapping. Tapping may be with whole hand or with a finger.

6. Contact point

Child touches an object with the index finger.

> i. The <u>*index or middle finger*</u> *must be extended, must touch the referent, and must be separated from the adjacent fingers. The thumb may remain close to the index finger; however, it should not be the primary digit contacting the indicated object.*
>
> ii. *At least two of the adjacent fingers should be curled under or arched up.*
>
> iii. When the child or adult is using the extended index finger to operate a toy (e.g., cash register buttons), this is not a contact point.

7. Abbreviated Reaching

Child reaches for an object but does not directly grab it. A reach must be *open-handed involving an extended arm and a momentary, expectant pause by the child*. The intention of the act may be imperative or declarative.

***8.* Moving an object away from the adult**

The child *distinctly and purposefully* moves the object away from the adult. *This is not the same as taking an object from the adult. Attention to adult must occur. Takes should not be coded as gestures.

9. Pantomime

Pantomime is the use of a part of the body or face to imitate an object or the use of an object. NO OBJECT IS USED IN PLACE OF ACTUAL OBJECT. For example:

i. Pretending to brush one's hair without a hairbrush

ii. Moving arms in a "rocking baby" movement without a doll

iii. Finger plays such as "Here's the church, here's the steeple..."

iv. Blowing a kiss.

10. Raising arms up

Child raises his or her arms up to indicate that he or she would like to be picked up. This act may be initiated by the adult.

11. Moving object toward adult

The move must be *discreet and purposeful* and meet the following requirements:

i. When seated across from each other, the child must move the object across the midline of the table. If seated in any other arrangement, the object must be moved at least half the distance between the two. The object should NOT be in the adult's hands at the time of the movement.

ii. The child *does not need to release the object* if the move is considered an "offer" or "rejection" that is not received by the adult listener.

iii. If the child begins to move the object to the adult then changes his mind, **this is not coded.**

iv. Brushing adult's hair is not coded as move object toward adult.

Examples:

Codeable Example: The child pushes his bowl of cereal at least halfway across the table in the direction of the adult to indicate "rejection".

Non-Codeable Example: The child pretends to pour tea in the adult's cup by moving the pitcher to the cup in the adult's hand without any expectation that the adult perform any action. This *does not* count as move toward the adult. The child would have moved the pitcher to the cup even if the adult were not holding it.

Non-Codeable Example: A "back and forth" ball game without vocalizations. Just taking a turn with a ball is not codeable.

12. Throwing or dropping an object

Remember that this act must have meaning in order to be coded as a gesture. Back and forth ball games should not be coded as gestures.

i. Knocking an object off the table can be coded as a gesture only when preceded by visual attention to the object being knocked off, and attention must be paid to the adult. (Act should be intentional and communicative)

ii. The drop occurs when the hand is not visible to the coder, code if the child has been dropping objects in the immediately preceding child acts or is looking where he is dropping the object before he drops it.

iii. If the drop is not visible, confirm the drop by listening for the sound of the object hitting the floor or by using the adult's following actions.

13. Wave

Child waves the hand or arm, can be directed towards an object or a person (for the purpose of greeting or saying goodbye).

14. Shrug of shoulders

A shrug includes lifting of the shoulders to the ears or upturning of palms to indicate "what" or "I don't know."

15. Upturned palm

The palm should be upturned as if to say "give that to me." There should be an expectant pause in which the child waits for the adult to react. The upturned palm must not be part of an act designed to retrieve an object independently.

16. Both palms upturned

Both hands are held palms up and to the side as if to say/ask “Where is ___?”

17. Come here

Upturned palm with one or more fingers or the whole hand wiggling to convey the message to come.

18. Shh sign

The sign must be distinctive with the finger held in close approximation to the mouth.

19. Head nod or shake

A head nod or shake must be intended to convey the message "yes" or "no."

20. Crossed arms

Child crosses arms over chest to show dissatisfaction not just a resting stance.

21. Clapping

Child brings hands together quickly in midline, palms usually touch briefly.

22. Hands over mouth
Child places one or both hands over mouth as if to express surprise.

23. Patting on a chair
Child pats chair, as in “sit by me”

24. Pushing object away

This is different from moving an object toward an adult in that the object may be in the adult’s hands. The purpose of this gesture is always rejection or a signal that the child is finished with the object. The object need not be pushed towards the adult’s body. For example, the child may knock the toy out of the adult’s hands or push the object (while still in the adult’s hands) towards the side. If the adult immediately pulls the object away, contact with the object may be brief. If no contact with the OBJECT is made, do not code as pushing away. Any portion of the child’s hand or arm may be used in pushing away.

25. Not otherwise specified

Gestures that are not on the list but appear to be clearly communicative may be coded as NOS.

26. No gesture performed

Occasionally, the coder may disagree with the original coding of a gesture. In this case, “No gesture performed” should be coded. This code should also be used with all other previously coded communication acts to ensure inclusion of the data file in analysis.

> **NOTE:** Dancing or moving to music (without touching) is **not** a gesture, even if the child and mom are looking at each other.