Published in final edited form as:
*Am J Speech Lang Pathol*. 2014 May 1; 23(2): 146–159. doi:10.1044/2013_AJSLP-13-0046.

# The Role of Maternal Gesture Use in Speech Use by Children with Fragile X Syndrome

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## Abstract

**Purpose—**The purpose of this study was to investigate how maternal gesture relates to speech production by children with fragile X syndrome (FXS).

**Method—**Participants were 27 young children with FXS (23 boys, 4 girls) and their mothers. Videotaped home observations were conducted between the ages of 25 and 37 months (toddler period), and again between the ages of 60 and 71 months (child period). The videos were later coded for types of maternal utterances and maternal gestures that preceded child speech productions. Children were also assessed with the Mullen Scales of Early Learning at both ages

**Results—**Maternal gesture use in the toddler period was positively related to expressive language scores at both age periods, and was related to receptive language scores in the child period. Maternal proximal pointing, in comparison to other gestures, evoked more speech responses from children during the mother-child interactions particularly when combined with wh-questions.

**Conclusion—**This study adds to the growing body of research on the importance of contextual variables, such as maternal gestures, in child language development. Parental gesture use may be an easily added ingredient to parent-focused early language intervention programs.

### Keywords

fragile X syndrome; gesture; language development; speech

Fragile X syndrome (FXS) is the most common inherited cause of developmental disability (Crawford, Acuna, & Sherman, 2001; Sherman, Morton, Jacobs, & Turner, 1984; Turner, Webb, Wake, & Robinson, 1996), affecting approximately 1 in 4,000 males and 1 in 6,000 females (CDC, 2011). FXS is caused by a mutation of the FMR1 gene located on the X chromosome (Verkerk et al., 1991) resulting in a wide range of disability that extends from learning disabilities to severe intellectual disabilities (Loesch et al., 2004). Because FXS is an X-linked neurogenetic disorder, males are usually more affected than females (Abbeduto, Brady, & Kover, 2007; Hagerman, 2007). Past research has estimated that co-occurring

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autism is present in as many as 30% of individuals with FXS (Denmark, Feldman, & Holden, 2003; Kaufmann et al., 2004; Philofsky, Hepburn, Hayes, Hagerman, & Rogers, 2004).

In the present study, we examine child speech responses to maternal gesture use in children with FXS. Specifically, we explored whether children with FXS were more likely to respond with speech to maternal utterances that included different types of gestures. Research on maternal gesture use and child language development has indicated that maternal gesture use helps to facilitate language development (Goodwyn, Acredolo, & Brown, 2000; Iverson, Capirci, Longobardi, & Caselli, 1999; McGregor & Capone, 2001; Namy, Acredolo, & Goodwyn, 2000; Rowe & Goldin-Meadow, 2009; Zammit, & Schafer, 2011), which in turn increases the child's gestural repertoire. Having a larger gestural repertoire and using gestures frequently in early language development helps children to reach language milestones earlier (Bates, Camaioni, & Volterra, 1975; Capirci et al., 1996; Folven & Bonvillian, 1991; Goodwyn et al., 2000; Morford & Goldin-Meadow, 1992). However, these previous studies focused on typically developing children, and did not examine children's immediate responses to maternal gestures.

## Language Delays in Young Children with FXS

Language development in most children with FXS is delayed (Abbeduto & Hagerman, 1997; Roberts, Mirrett, Anderson, Burchinal, & Neebe, 2002; Roberts, Mirrett, & Burchinal, 2001). Research on both expressive and receptive language in young children with FXS indicates significant delays in both domains by 18 months (Hatton et al., 2009; Mirrett, Bailey, Roberts, & Hatton, 2004; Roberts et al., 2009). It appears that more pronounced delays are evident as early as 9 months in expressive language (Roberts, Mirrett, et al., 2001) and this pattern appears to continue over the course of language development (Roberts et al., 2009). In addition, the average age of first words in boys with FXS is 28 months (range 9–88 months), which highlights the severity of expressive language delays in this population (Roberts, Hatton, & Bailey, 2001). Receptive language, on the other hand, appears to develop more quickly in children with FXS, albeit delayed when compared to typically developing children (Roberts, Mirrett, et al., 2001).

Children with FXS tend to remain in the prelinguistic period of language development longer than typically developing children (Brady, Skinner, Roberts, & Hennon, 2006). Similar to children with other intellectual and developmental disabilities (IDD; Abrahamsen, Cavallo, & McCluer, 1985; Caselli et al., 1998; Chan & Iacono, 2001; Goodhart & Baron-Cohen, 1993; Singer Harris, Bellugi, Bates, Jones, & Rossen, 1997), prelinguistic communication may be an effective form of communication for children with FXS. However, Flenthrope and Brady (2010) found that children with FXS and higher levels autism symptoms who produced more communicative gestures had lower vocabularies two years later, which is the opposite of what has been observed in typically developing children and children with IDD. More research on the critical transitional period from prelinguistic to linguistic communication in FXS is needed.

Fostering prelinguistic communication through early language interventions can help to promote communication and language development in children with FXS (Brady, Bredin-Oja, & Warren, 2007); however, the ultimate goal for families is for their children to communicate with speech (Brady et al., 2006). This goal is understandable because, although all forms of communication are important, speech is the most efficient and most widely recognized form of communication. In addition to parents, educators and society view the onset of speech as an important developmental milestone. For example, psychoeducational tests of young children frequently include the age of first words as an important indicator of language development. Also, communication partners respond differently to speaking versus nonspeaking children (Beck & Fritz-Verticchio, 2003; Soto, 1997). Therefore, identifying variables that contribute to speech development in children with FXS is of the upmost importance.

## Importance of Parental Gesture Use in Language Development

Adults frequently use gestures in addition to speech when interacting with infants and young children, which appears to help reinforce the spoken message (Iverson, Capirci, Longobardi, & Cristina Caselli, 1999; McNeil, Alibali, & Evans, 2000; O'Neill, Bard, Linnell, & Fluck, 2005). The type of gestures adults use in their communicative interactions with infants and young children is a modified form of adult gestures (O'Neill et al., 2005; Shatz, 1982). This simplified, context based, and concrete form of gesture use that parents, and adults, use when interacting with their children has been termed "gesturese" because these child-directed gestures are the gestural equivalent of child-directed speech or "motherese" (Iverson et al., 1999; O'Neill et al., 2005; Schmidt, 1996; Shatz, 1982). Theoretically, added support from gestures allows children to comprehend speech at a higher level than if only a spoken message was provided. The use of gestures as a scaffold for communication begins in infancy as infants shift their attention to the world around them and continues into childhood (Trevarthen & Aitken, 2001). During this time, parents use gesturese and vocalizations to help with initiating and maintaining infant and child attention (O'Neill et al., 2005; Tomasello & Farrar, 1986).

Research on typically developing children, preterm infants, and children with Down syndrome (DS) has noted that children increase their attention to objects when parents reference them simultaneously with both gestures and speech (Landry & Chapieski, 1989; Legerstee, Vargehese, & Van Beek, 2002; Roach, Barratt, Miller, & Leavitt, 1998). Because gestures help to reinforce the spoken word or message, these interactions are important for language comprehension and promote language learning (Akhtar, Dunham, & Dunham, 1991; Legerstee et al., 2002; Saxon, Frick, & Colombo, 1997; Tomasello & Farrar, 1986). Further, there is evidence that using gestures with speech to help maintain child attention to objects increases the likelihood of children with DS responding to adult initiations (Landry & Chapieski, 1989; Legerstee et al., 2002; Roach et al., 1998). Specifically, Legerstee and colleagues (2002) found that the use of verbal and nonverbal techniques to maintain infant attention by mothers of children with and without DS promoted the use of gestures and words by infants. Research by Roach and colleagues (1998) on infants with DS, a group of mental age matched infants, and chronological age matched infants found that more vocalizations and object play by children with DS was associated with more supportive maternal behaviors.

Taken together, gestures appear to scaffold speech comprehension and the achievement of language and other milestones in young children with and without developmental disabilities (Capone & McGregor, 2004; McNeil et al., 2000; Ninio & Bruner, 1978; Ratner & Bruner, 1978; Vygotsky, 1978; Wood, Bruner, & Ross, 1976). The link between parental gesture use and later language development may be especially important for promoting communication in children with FXS and IDD because of the delayed onset of speech associated with these disorders (Abbeduto et al., 2007; Brady, Marquis, Fleming, & McLean, 2004; McLean, McLean, Brady, & Etter, 1991). A strong visual link between referent and word could help bridge the transition from child gestural communication to spoken communication.

Parents of children with IDD may use gestures more frequently in their communicative interactions with their children (Chan & Iacono, 2001). This prolonged gesture use by mothers of children with IDD may help to make information more accessible to children by adding additional scaffolding (Capone & McGregor, 2004). Also, continuing to use gestures in later periods of development may be one technique mothers use to adapt their communication style to their child’s level of communication and ability (Capone & McGregor, 2004; Iverson, Longobardi, Spampinato, & Cristina Caselli, 2006). For example, research on mothers of children with DS by Iverson and colleagues (2006) indicates that mothers of children, who have a greater lag in expressive language skills, produce more utterances that include gestures than mothers whose child had a lesser lag. Further, it seems that continuing to pair gestures with spoken messages later in development may help to facilitate effective communication between mothers and children with DS (Iverson et al., 2006; Wang, Bernas, & Eberhard, 2001).

Considering the wealth of information demonstrating the beneficial relationship between maternal gesture use and later language development, it is surprising that more early childhood research has not focused on observed measures of child speech responses immediately following maternal gestures. As hypothesized by Iverson and colleagues (1999), maternal gesture use may help to promote vocabulary development in children because gestures “can single out a referent from other objects in the context and make its relation to accompanying speech more salient” (p. 60), which provides the child with more supporting information about the word. Iverson and colleagues (1999) showed that, for typically developing children, the frequency of maternal gesture use at 16 months significantly predicted child word production and gesture use at 20 months (Iverson et al., 1999). Similarly, Talbott, Nelson, and Tager-Flusberg (2013) found that maternal gesture use at 12 months was positively correlated with both infant gesture use and language ability at 18 months for infants who were at a low risk for autism (i.e., typically developing infants with no family history of autism) and for those infants that were at a high risk for autism (i.e., has an older siblings with autism; Talbot et al., 2013).

One reason that there has not been more research on gesture use by mothers is that conducting this type of research presents many challenges. Gestures are visual observations and, therefore, must either be examined live or through videotape analyses, requiring more

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resources to collect primary and reliability data. In addition, although there is general agreement about the forms of conventional gestures (Bakeman & Adamson, 1986), there is extensive variability between gesture productions that can impede coding accuracy.

### Pointing gestures

One type of gesture that appears to be particularly important for communication is deictic gestures, which are used to identify the presence of an object, person, or event (Iverson et al., 1999; O'Neill et al., 2005). Deictic gestures include pointing, showing, giving, and other gestures that are used to indicate the object or event that is the focus of the communicative interaction, such as tapping or touching an object (Bates, 1976; Iverson et al., 1999; Masur, 1990; O'Neill et al., 2005; Tfouni & Klatzky, 1983; Zammit & Schafer, 2011). The most common and important deictic gestures for communication are pointing gestures (Iverson & Goldin-Meadow, 2005; Ozçalişkan & Goldin-Meadow, 2005; Tomasello, Carpenter, & Liszkowski, 2007; Tomasello, 1988) because a pointing gesture can help to direct child attention to the object or event that is the focus of the communicative interaction (Bates et al., 1975), thus, helping to maintain infant and child attention (Bangerter, 2004; Liszkowski, Carpenter, Henning, Striano, & Tomasello, 2004; Louwerse & Bangerter, 2010; Tomasello et al., 2007). Further, pointing gestures are the most common gestures used by mothers during mother-child interactions (Iverson et al., 1999; O'Neill et al., 2005). Maternal pointing gestures help promote children's understanding of speech because they help to clarify the verbal utterance (O'Neill et al., 2005) as evidenced by research on different aspects of child language comprehension (i.e., message comprehension [Schmidt, 1996], word comprehension [Morford & Goldin-Meadow, 1992; Tfouni & Klatzky, 1983], and acquisition of nouns [Gogate, Bahrick, & Watson, 2000; Moore, Angelopoulos, & Bennett, 1999; Schmidt, 1996; Zammit & Schafer, 2011]).

## Present Study

The purpose of the present study was to investigate children's speech responses following maternal utterances that included gesture use as part of their child-directed communication acts. This investigation focused on children's speech responses to maternal gesture use between the ages of 24 and 37 months (toddler period) and again between 60 and 71 months (child period). This allowed us to examine the influence of different maternal gestures early in speech development – when children with FXS may not be speaking much and maternal gestures may be most important – versus later in speech development – when children are speaking much more and maternal gestures may play a less critical role. The following research questions were addressed:

1. Does maternal gesture use during the toddler and child period relate to receptive and expressive language development in children with FXS?
2. Do children with FXS speak more following maternal utterances that contain specific gesture types in both the toddler and childhood periods?
3. Is the association of child speech with maternal gesture stronger during the toddler period, as compared to the child period?

**a.** Is child speech more likely to be preceded by pointing than other types of gestures?

**b.** Is differential responding to pointing more pronounced in the toddler or the child period?

# Method

## Participants

Participants were 27 young children with FXS (23 boys, 4 girls) and their mothers. Participants were chosen from a sample of families participating in a larger, longitudinal study of family adaptation in FXS (see Warren, Brady, Sterling, Fleming, & Marquis, 2010 for details). For this study, we examined data from all families in the larger study who had video data when the child was between 25 and 37 months (Time 1) and again when the child was between 60 and 71 months (Time 2; see Table 1 for descriptive information). These two age periods will be referred to as *toddler* and *child.* The mean age at the toddler period was 32.67 months (SD = 3.44) and the mean age at the child period was 66.37 months (SD = 3.03). Families were recruited from across the United States through networking with FXS family support groups, using a national research registry, and advertising at national conventions and on an FXS parent listserve. The sample represents families from 24 of the 50 states. The median household income was $70,000 (range $32,000 – $250,000). All of the mothers that had been tested for FXS were premutation carriers (two of the mothers had not been tested).

Table 1 presents descriptive information for the child participants and their mothers. The Early Learning Composite and age equivalent scores for the receptive and expressive language domain (Mullen, 1995) at both age periods are presented to provide information on the overall developmental level and functioning of the children. Although 6 of the children were above the age of 68 months (the age cut off of the MSEL) during the second observation, we still administered the MSEL to these children and we report the composite and age equivalent scores associated with 68 months. Similarly, the Childhood Autism Rating Scale (CARS; Schopler, Reichler, & Renner, 1988) is presented to provide information on the level of autism symptomology at both age periods, which could also influence the overall functioning of the child. A CARS score below 30 means that the child has low or no autism symptoms, a score from 30 to 36 is in the mild to moderate range for autism symptoms, and a score above 36 is an indicator of high autism symptoms. The CARS score was not used as a diagnostic indicator, but simply for descriptive purposes in this study.

## Measures

**Mullen Scales of Early Learning—**The Mullen Scales of Early Learning (MSEL; Mullen, 1995) is a standardized observational measure of development for children between the ages of 3 to 68 months. There are five domains of the MSEL (i.e., gross motor, fine motor, visual reception, expressive language, and receptive language) and an overall score (i.e., Early Learning Composite) that provides an estimate of overall developmental functioning. Strong concurrent validity for the MSEL has ben established with other developmental assessments for young children (e.g., Bayley Scales of Infant Development [Bayley, 1993], Birth to Three Scales [Dodson & Bangs, 1979], Peabody Developmental Motor Scales [Folio & Fewell, 1983]). The MSEL has strong test-retest reliability coefficients (.82-.85), internal consistency coefficients (.83-.93), and interrater reliability (.91-.99). Also, content, construct, and predicate validity have been established for the MSEL (Mullen, 1995).

## Procedure

**Data collection and contexts—**Information about maternal gesture use, maternal utterance, and child speech responses was obtained from observational video data at both age periods. During the observation, mother-child dyads interacted during a series of structured and unstructured contexts. The structured interactional contexts involved reading a book together, eating a snack together, and a free play session. These contexts were classified as structured because the dyads were provided with the materials (i.e., books to read, snacks, free play toys) to complete these activities. However, the mothers were told that they could complete these activities at any location in their home and to interact as they normally would. Each of the structured interactional contexts was five minutes long. The full five-minutes of each of these contexts were used for coding purposes. The unstructured interactional context was a 30-minute naturalistic observation. For the naturalistic observation, mothers were asked to interact with their child as they normally would during daily activities (i.e., playing together, doing chores, etc.). The only request made of mothers was that they stay in the same room as the child and not turn on the television. Two (2) five minute samples were selected from this 30-minute observation for coding purposes. The samples taken from the video data for coding purposes occurred between minutes 5 to 10 and minutes 20 to 25 of the observation. The first sample time of 5 to 10 minutes was chosen because it was toward the beginning of the session, but after 5 minutes of “warm up time,” and the second sample from 20 to 25 minutes was chosen because we thought that by the time 20 minutes had elapsed, mothers might be use to the presence of the observers and hence act more naturally. Thus, a total of 25-minutes sampled across the following contexts were scored for the present analyses: book reading, snack, free play, and naturalistic.

**Coding—**Video files were digitized for coding using Noldus™ Observer software, version 5.1 (Noldus, 2002). The second author and three (3) trained speech-language graduate students served as coders for this study. Once trained, the graduate students acted as the primary coders and the second author served as the reliability coder. Primary coders were trained to search the previously coded and time stamped transcripts from Noldus™ Observer for the code of *maternal gesture*. Once located, the maternal gesture type and child response type were coded by indicating the corresponding code number for the behavior on the printed transcript (see Table 2).

Maternal gestures that were part of communication acts directed to their child with FXS were identified and coded with one of the codes (affection, proximal point, nod/shrug, give/take, distal point, and other) listed in Table 2. A code of *other* for maternal gestures was used if a gesture did not fit into one of these categories. Thus the coding scheme was exclusive and exhaustive. Table 3 provides the means and standard deviations for the maternal gestures performed at the toddler and child periods. In addition, once the maternal gesture was identified, the maternal utterance that occurred with the gesture was transcribed and subsequently coded with one of the codes (wh-question, directive, yes/no question, label, and other) listed in Table 2. A code of *other* for maternal utterance was used if a maternal utterance did not fit into one of these categories.

Child responses to each of these maternal communication gestures were coded as one of the following: gesture, gesture and speech, speech only, or other (see Table 2 for definitions of child response codes). The code *other* for the child was used when the child did not respond to maternal gestures or responded with a non-communicative behavior such as behavioral compliance to a maternal request (e.g., sitting down when asked to do so).

**Reliability:** Forty percent of these videos were randomly selected for independent reliability coding. We calculated Cohen's *kappa* for each maternal gesture, maternal utterance, and child response code. The magnitude of kappa represents the proportion of agreement greater than that expected by chance and coefficients between .61 and .80 are viewed as "substantial" (Landis & Koch, 1977). The mean kappa obtained across the videos was .77 and the range of coefficients was .52–.90.

**Data reduction—**Because we were interested in child speech responses to maternal gestures, the child responses coded as *speech only* and *gesture and speech* were collapsed into a new variable called *any speech response*. Similarly, the child responses coded as *gesture* and *other* were collapsed into a new variable called *other response.* This allowed for an examination of *speech* as opposed to *non-speech* responses during analyses.

Maternal gesture categories were also collapsed. Mothers performed the maternal gesture codes of affection, nod/shrug, give/take, distal point, and other at low rates (10% or less of all mother utterances -- Table 3 for details). The low rates of these gestures mean that it was not possible to statistically explore the role of each of these gestures individually in our analysis. Therefore, those four maternal gestures were collapsed into a new variable called *any other gesture*. For data analysis, the maternal gesture types were *proximal point* and *any other gesture*. Similarly, the maternal utterances were coded as yes/no question, directive, label, and other also occurred at low rates (these utterances were observed in 15% or less of all mothers --Table 3 for details). Because nearly half of the utterances preceding child speech were wh-questions, the maternal utterance codes were collapsed into two variables for analysis: *wh-question* and *any other speech.*

**Contexts:** Although mother-child interactions were coded in multiple contexts, descriptive examination of maternal gesture use in the five contexts indicated that the gesture use in each context was similar (see Table 4). Therefore, we did not perform additional analyses to determine if children with FXS were more likely to respond with speech to maternal gesture use in different contexts. That is, we analyzed maternal and child responses across all five contexts combined.

**Analysis plan—**To examine the first research question of whether maternal gesture use during the toddler and child period related to receptive and expressive language development, we used Spearman rho correlations. It was necessary to use Spearman rho correlations instead of Pearson *r* correlations because the variables of maternal gesture use at the toddler and child period were skewed and did not have a normal distribution, thus violating the assumptions of the Pearson *r* correlation. In order to examine the role of maternal gesture use on child receptive and expressive language abilities as measured by the MSEL, the raw scores for these domains were used for analysis. Raw scores provided a more accurate measure of growth in these skills as compared to the T-scores, which are influenced by the child's chronological age. In addition, raw scores were more appropriate for the 6 children who were older than 68 months.

In order to address the second research question about the relationship between maternal gesture use and child speech responses, we analyzed data from mother-child dyads at each age period. Each of these observations contained many maternal gestures, each paired with an opportunity for child response (i.e., within 3 seconds after the mothers gesture). This yielded nested data wherein the maternal gestures and child responses were nested within each mother-child dyad, creating dependency between observations. Each dyad consisted of a mother-child pair, with many features unique to itself, including not only demographic characteristics, but also the dyad's history of interactions over the course of the child's life. Using multi-level modeling allows for the examination of the effects of maternal gesture type, maternal utterance, and age period on child speech, after taking into account the effect of dyad. Also, each dyad varied in the number of observed gestures and each child varied in the amount of speech they used. Therefore, common assumptions (i.e., independence and homogeneity of variance) of many alternate analyses were not met. Specifically, the variables were not independent because each child response was tied to the gesture performed by the mothers. Finally, each dyad had a different number of interactions making them unbalanced designs with different variances between groups. Multi-level modeling is ideal for examining data in situations involving repeated measures, as is the case in this study, due to its ability to handle complicated models including those with missing data and unbalanced designs (Misangyi, LePine, Algina & Goeddeke, 2006).

We used Odds Ratios to present the results because the outcome, presence or absence of a child's speech response, was dichotomous. Odds Ratios are a method of demonstrating the strength of the association between a predictor and a dichotomous outcome of interest (UCLA, 2013; Watson, Crais, Baranek, Dykstra & Wilson, 2013). For the purpose of this study, the Odds Ratio represents the probability of a particular gesture being associated with a speech response. Odds Ratios above 1 indicated that the event (maternal gesture type) is more associated with child speech, while Odds Ratios below 1 indicated that the event is more associated with the absence of speech.

As previously noted, many maternal utterances that accompanied gestures were wh-questions. In order to examine how wh-questions influenced child speech, the multi-level analysis was repeated for the subset of maternal gestures that were paired with wh-questions, as well as those paired with other utterances.

For the examination of the third research question about whether the association between child speech and maternal gesture was stronger during the toddler period, as compared to the child period, we used the same modeling approach separately for each of the age periods. In addition, the evocative effect of maternal proximal pointing was explored in this model.

## Results

### Relationship between Maternal Gesture Use and Child Language Outcomes

Spearman rho correlations were performed to examine the first research question about the relationship between maternal gesture use and raw scores on receptive and expressive subtests from the MSEL at both age periods. Results of these analyses indicated that there was a positive correlation between maternal gesture use and expressive language abilities, $r_s(27) = .59$, $p < .001$, for the toddler period, such that children with FXS whose mothers used more gestures had higher expressive language scores on the MSEL. However, no statistically significant relationship was observed between maternal gesture use and receptive language abilities, $r_s(27) = .34$, $p = .08$ for the toddler period. Also, no statistically significant relationship was observed between maternal gesture use and either receptive, $r_s(27) = .28$, $p = .16$, or expressive language abilities, $r_s(27) = .23$, $p = .26$, for the child period.

Correlations were also performed to examine the relationship between maternal gesture use during the toddler period and receptive and expressive language abilities measured during the child period. The results of these analyses indicated a statistically significant relationship between maternal gesture in the toddler period and both expressive language, $r_s(27) = .50$, $p = .008$, and receptive language abilities, $r_s(27) = .44$, $p = .02$, in the child period. Specifically, children with FXS whose mothers used more gestures during the toddler period had higher expressive and receptive language scores on the MSEL during the child period.

### Child Speech in Response to Maternal Gesture Use

In order to address the second research question about child speech in response to maternal utterances containing gestures, a multilevel model using SAS PROC GLIMMIX was used to model speech at each age period. Fixed effects terms for gesture type, at level 1, and dyad, at level 2, were included. The same model was then performed separately for the two types of maternal utterance (wh-questions and any other utterance), and then with each of these utterances types combined with the maternal gesture types (proximal point and any other gesture) with child speech as the outcome variable. The model was also used for both the toddler and child period to examine research question three about whether the association between child speech and maternal gesture was stronger during the toddler period, as compared to the child period. Descriptive information about the numbers of each type of maternal gesture is provided in Table 3.

**Toddler period—**Results from the multilevel model analyses are presented in Table 5. During the toddler period, child speech was strongly related to maternal gesture use, $F(1,1063) = 11.03$, $p = .0009$. Specifically, results indicated a positive relationship between proximal pointing and child speech, as demonstrated by the positive values of the estimated slopes in Table 5. In the first row, the effect of the maternal gesture *proximal point* is contrasted with *any other* types of gestures. The positive value of the Estimated Slope indicated a positive relationship between the *proximal point* gesture and child speech. Because the slope was large compared to the standard error (SE), and the *p* value was small ($p = .0009$), the effect of *proximal point* on speech can be interpreted as significant.

The Odds Ratios (OR) shows the relative likelihood of each gesture in each comparison being associated with child speech. As can be seen in table 5 during the toddler period, child speech was almost twice as likely to follow a *proximal point* than any other type of gesture as indicated by odds ratio of 1.95, which can be seen in the OR Estimate column (see Table 5).

**Child period—**During the child period, child speech was also strongly related to maternal gesture use, $F(1, 1152) = 86.40$, $p < .0001$. In fact, contrary to our initial hypothesis, the relationship between child speech and maternal gesture use was actually stronger during the child period than during the toddler period. This can be seen not only in the magnitude of *F*-value, but also in the size of the estimated slopes and OR. Specifically, child speech was nearly four times more likely to follow a maternal *proximal point* than any other type of maternal gesture during the child period as indicated by an OR of 3.94 (see Table 5).

**Combined child speech response to maternal gestures:** In order to address the third question comparing the evocative effect of gestures across the toddler and child periods, data from both age periods were included in a single model and the model used in the prior analyses was adapted slightly. For the combined analysis, an additional fixed effects term for time period was included at level 1, along with the effects for gesture type (level 1) and dyad (level 2). When the two time periods are analyzed together, both maternal gesture type, $F(1, 2216) = 120.72$, $p < .0001$, and age period, $F(1, 2216) = 239.23$, $p < .0001$, were significant predictors of child speech. As expected based on the first two models, child speech following any gesture was more than five times as likely to occur during the child period than during the toddler period as shown by an OR of 5.26. As in the individual models for the toddler and child period, maternal proximal pointing was positively associated with child speech. Child speech was approximately three times more likely to occur following a proximal point than any other gesture, as shown by an OR of 3.13 (see Table 5). Preliminary analysis indicated that there was no interaction between age period and maternal gesture type, so this effect was dropped from the final model.

***Role of maternal utterance on the effect of proximal pointing:*** Because of the frequent occurrence of maternal wh-questions, maternal utterances containing *wh-questions* were examined separately in a post-hoc analysis. Within maternal *wh-question* utterances, both maternal gesture type, $F(1, 470) = 24.42$, $p < .0001$, and age period, $F(1, 470) = 36.84$, $p < .0001$, were significant predictors of child speech. Utterances that combined *wh-questions* with *proximal point* gestures were nearly five times as likely to precede child speech than were maternal utterances that combined wh-questions with other gestures as shown by an OR of 4.93 (see Table 5).

When only those utterances *not* containing a wh-question were examined, both maternal gesture type, $F(1, 1743) = 38.42$, $p < .0001$, and age period, $F(1, 1743) = 151.93$, $p < .0001$, were significant predictors of child speech. Proximal pointing continued to be a significant maternal gesture for promoting child speech, even when not combined with a wh-question utterance. Specifically, children were twice as likely to respond with speech following a non wh-question utterance combined with a proximal point than to a non wh-question utterance combined with another gesture.

In order to further examine the influence of both maternal proximal pointing and wh-questions on child speech, we created a variable that combined these two constructs. When *proximal point* and *wh-questions* were combined into a single variable, they significantly predicted child speech, $F(1, 2215) = 103.30$, $p < .0001$, and age period, $F(1, 2215) = 195.67$, $p < .0001$. Children were 11.24 times more likely to respond with speech when mothers used both a proximal point and a wh-question. Thus, when mothers used either a proximal point or wh-question children were not as likely to respond with speech, as they were when mothers used a proximal pointing gesture with a wh-question (see Table 5).

**Summary—**Taken together, the results of these models indicate that there was an evocative effect of gesture on speech across the two age periods. Children with FXS – regardless of the age period, or the content of the maternal utterance paired with the gesture – were more likely to respond with speech following a maternal *proximal pointing* than any other gesture. This effect was most pronounced when proximal pointing was combined with *wh-questions*, but also pertained to maternal utterances that did not contain *wh-questions*. It is important to note that for our analysis a total of 10 statistical tests were conducted, so it was necessary to adjust the alpha level for these multiple comparisons. Accordingly, to control for family-wise error rate we used the Holm-Bonferroni Method in which the most significant effect is compared to a *p*-value of alpha divided by the number of tests (i.e., $\alpha/10 = .005$), with each subsequent test being compared to a larger *p*-value (i.e., $\alpha/9$, $\alpha/8$, …, $\alpha/2$, $\alpha$). Because all the tests were significant below an alpha of .001, we can be confident that the significance level is not due type 1 error.

## Discussion

The purpose of the present study was to examine the relationship between maternal gestures and language in children with FXS. We reported results for two child language outcomes – the raw scores of receptive and expressive language from the MSEL and child speech responses during mother-child interactions. Specifically, we examined whether children were more likely to respond with speech following different maternal gesture types. Past research has suggested that maternal gesture use helps to promote language outcomes (Goodwyn et al., 2000; Namy et al., 2000; Zammit & Schafer, 2011), and that children are more likely to respond with gestures and speech when mothers use gestures during communicative interactions (Iverson et al., 1999). Using gestures appears to help scaffold communicative interactions by providing children with more information about the spoken message, which in turn promotes comprehension (Capone & McGregor, 2004; Iverson et al., 1999; McNeil et al., 2000; Ninio & Bruner, 1978; Vygotsky, 1978).

A positive relationship was only observed between maternal gesture use and expressive language raw scores for children with FXS within the toddler period. Therefore, this study supports the notion that maternal gesture use, at least early in development, is associated with higher expressive language scores in children with FXS. Further, maternal gesture use in the toddler period was positively related to both expressive and receptive language scores in the child period. These findings add further support to the notion that maternal gesture use early in development is associated with later language development. However, the relationship between concurrent receptive and expressive language scores and maternal gestures during the child period was not significant. One possible explanation for this is that use of maternal gestures related to the earlier items on the MSEL receptive and expressive scales that were reflected during the toddler period, but gestures did not specifically relate to more complex language behaviors measured during the child period.

The second set of analyses looked at contingent responses of children to their mothers during dyadic interactions. By focusing on the maternal antecedents to children's speech, it was possible to determine if specific maternal gestures were more likely to evoke children's speech responses. The use of multi-level modeling allowed for the discovery that, regardless of age period, maternal proximal pointing was most likely to evoke a speech response from the child. In our observations, nearly all maternal pointing was proximal as opposed to distal pointing. Low rates of maternal distal points may have been observed because the video data for this study was designed to examine mother-child interactions within relatively close proximity to each other. Because of this, mothers may have been focusing on face-to-face interactions with their child and not directing their child's attention to objects and events outside of the interaction.

The results of the present study indicate a potential skill to target for intervention-- proximal pointing gestures by mothers. Most of the research on pointing has focused on the importance of distal points for promoting communication (see Kita, 2003 for a review), it appears, however, that proximal points are also important for communication. Proximal points may be particularly salient for promoting communication because they directly link referents to their verbal mappings, and may also help to clarify a verbal utterance (O'Neill et al., 2005). For example, when a mother proximally points to a picture of a "kitty" while saying "kitty" the relationship between word and referent is clearer than when a similar utterance is not accompanied by a point. These findings suggest that proximal pointing, in comparison to other gestures, evokes more speech responses from children with FXS, and that this is particularly true in the childhood period. In addition, it appears that the combination of a proximal pointing gesture along with a wh-question (i.e., saying, "What's that?" while pointing to an object) is particularly effective in leading children to respond with speech.

Numerous studies have shown a strong relationship between parental input and child communication development (Fernald, Marchman, & Hurtado, 2008; Hart, 1991). Based on this research, numerous interventions have been developed to increase specific inputs to children with language delays (Buschmann et al., 2009; Drew et al., 2002; Girolametto & Wieitzman, 2006). Within these programs, parents are encouraged to scaffold children's responses and this scaffolding often includes adding gestures to child-directed communication. Our findings suggest that encouragement of proximal pointing may be a specific strategy to promote speech development. However, the results of this study are correlational and should be interpreted with caution. More research is needed to examine the influence of maternal proximal points on promoting language development and children's use of speech.

In addition to maternal proximal pointing gestures, maternal give, show, nod/shrug, affection, and distal point gestures were examined. Overall, children spoke less often following these maternal gestures than they did following proximal points. This may be because these gestures are more likely to produce an action or subsequent gesture from the child, rather than speech. For example, a mother may use a give gesture and the child responds by either taking or pushing away the item.

## Implications

The findings from the present study suggest that maternal gesture use, particularly proximal pointing, is an aspect of communication input that should be encouraged through intervention. Interventions designed to educate parents and caregivers about how to use gestures when communicating with their child could help to promote the child's developing communication skills, especially their vocabulary development and language ability, in contexts outside of the intervention and after the intervention has ended (Capone & McGregor, 2004; Ingersoll & Lalonde, 2010; Iverson & Goldin-Meadow, 2005; McGregor, Rohlfing, Bean, & Marschner, 2009; Stone, Ousley & Littleford, 1997; Stone & Yoder, 2001; Vaughan Van Hecke et al., 2007).

Although the current study focused on children with FXS, it is likely that similar results would be obtained for children with language delays associated with other types of IDD. Our findings lend additional support to parent-focused language intervention strategies that have been developed for children with other types of disabilities, and that include a gesture component. Interventions that aim to improve language by increasing parent responsivity (e.g., Fey et al., 2006; Landry, Smith, & Swank, 2006; McDuffie et al., 2013; Venker, McDuffie, Ellis Weismer, & Abbeduto, 2012) could be altered to focus more on maternal gestures. Specifically, adding proximal pointing to these interventions may be especially facilitative for child language development. More research that focuses on the development, implementation, and evaluation of early language interventions for children with IDD, including children with FXS, is needed. It is important to note that language interventions developed for children with FXS will also need to account for the influence of the FXS phenotype, especially behaviors that may subsequently interfere with their language development (i.e., challenging behaviors, attention, gaze aversion, social anxiety, etc.; Abbeduto & Chapman, 2005; Abbeduto et al., 2007; Hagerman, Rivera, & Hagerman, 2008; Sterling & Warren, 2008). Further, these behaviors may make it more difficult for parents to use responsive interaction strategies that facilitate communication (Abbeduto et al., 2007).

## Limitations

There are several limitations to the present study. First, the present sample was a sample of convenience and, therefore, is not representative of the population of children with FXS. Future research on gesture use in children with FXS should include a larger and more diverse sample. Results comparing maternal gestures to child language scores were correlational, and therefore cannot be interpreted as reflecting a causal connection between

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either maternal gestures or language. Also, it is possible that the present findings were influenced by the fact that mothers were being observed and videotaped while interacting with their children. This may have lead mothers to talk to their child more and/or use more gestures then they do under normal circumstances; thus, increasing both their amount of utterances and gesture use. Finally, only child speech responses to maternal gestures in children with FXS were examined. In order to understand whether this pattern of performance is similar to children in the same developmental period with different diagnoses, future studies should include children with other IDD and typically developing children.

### Conclusion

The findings from this study add to the growing body of research on communication in children with FXS. Also, this study adds to research on the importance of maternal gestures in child language development. Future studies should continue to examine the influence of maternal gestures on child speech production. Continuing to examine the relationship between maternal gesture use and child responses may have implications for the development of early language interventions for children with FXS and other developmental disabilities. Future research is needed to examine a possible causal relationship between maternal gesture use, particularly pointing gestures, and language development in children with FXS. Encouraging parental gesture use has the potential to be an important ingredient in parent-focused early language intervention programs.

## Acknowledgments

This research was supported by NICHD grant P30 HD02528, P30 HD003110, and T32 HD057844. We would like to thank Liz Morrey Korthanke, Juliana Keller, and Amy Green who assisted in data coding.

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## Table 1

### Participant Characteristics

(n = 27)

| Characteristic | | Time 1 | | | Time 2 | | |
|---|---|---|---|---|---|---|---|
| | % | M | SD | Range | M | SD | Range |
| Child | | | | | | | |
| Age (in months) | | 32.67 | 3.44 | 25–37 | 66.37 | 3.03 | 60–71 |
| Mullen Early Learning Composite | | 56.11 | 11.75 | 49–101 | 55.37 | 12.59 | 49–99 |
| MSEL Receptive Raw Score | | 19.96 | 6.64 | 4–33 | 33.63 | 8.15 | 17–48 |
| MSEL Receptive Age Equivalence (in months) | | 18.07 | 7.03 | 2–37 | 39.67 | 13.50 | 16–69 |
| MSEL Expressive Raw Score | | 16.81 | 7.00 | 5–33 | 30.63 | 9.73 | 11–47 |
| MSEL Expressive Age Equivalence (in months) | | 17.00 | 7.94 | 4–37 | 35.19 | 13.57 | 10–63 |
| CARS Score | | 25.22 | 5.72 | 15.5–35 | 25.17 | 5.91 | 16–36.5 |
| Gender (% male) | 85.2 | | | | | | |
| Maternal | | | | | | | |
| Age (in years) | | 32.44 | 4.25 | 25–41 | | | |
| IQ | | 110.22 | 13.76 | 73–130 | | | |
| Marital Status | | | | | | | |
| Married | 85.2 | | | | | | |
| Single | 7.4 | | | | | | |
| Divorced | 7.4 | | | | | | |
| Education | | | | | | | |
| High Diploma School or Less | 11.1 | | | | | | |
| Some College | 33.3 | | | | | | |
| College Degree | 25.9 | | | | | | |
| Some Post College | 3.7 | | | | | | |
| Master’s Degree or Advanced Degree | 25.9 | | | | | | |
| Racial/Ethnic Identity (%) | | | | | | | |
| Caucasian | 96.3 | | | | | | |
| Multi-Racial | 3.7 | | | | | | |

**Table 2**

Definitions of Maternal Gestures and Child Responses

| Category | Variable | Definition |
|---|---|---|
| Maternal Gestures | Affection | Tickling, clapping, waving, high five |
| | Proximal Point | Single extended finger towards a close object or event |
| | Nod/Shrug | Affirmative or negative head nods, unsure shrugs |
| | Give/Take | Give, take |
| | Distal Point | Single extend finger to object or event 6″ or more away (usually arm is extended) |
| | Other | Any gesture observed that does not fit into one of the above categories |
| | Any Other Gesture | New variable created by combining the variables of “affection”, “nod/shrug”, “give/take”, “distal point”, and “other” because of low rates of performance |
| Maternal Utterance | Wh-Question | Any what, when, where, why or how question directed to the child in anticipation of an open-ended response either gestural or verbal. Including any, “do you know” plus a wh-question (e.g., Do you know what his name is?) |
| | Yes/No Question | Any question that either requires or implies a request for a yes/no or either/or answer from the child (including tag questions) |
| | Directive | Any utterance requiring the child to perform or stop an act. This includes indirect directives. |
| | Label | Any utterance that represents the name, location or state of an item or person |
| | Other | Any utterance that does not fit into the categories. Including social greeting, praise or sympathy statements, reading, and sounds |
| | Any Other Speech | New variable created by combining the variables of “yes/no question”, “directive”, “label”, and “other” because of low rates of performance. |
| Child Responses | Gesture and Speech | Combination of gestures with any intelligible speech (i.e., any word that was recognizable) |
| | Speech Only | Intelligible speech (i.e., any word that was recognizable) |
| | Any Speech | New variable created by combining the variables “gesture and speech” and “speech only” for data analysis |
| | Gesture | Affection, proximal point, nod/shrug, give/take, distal point |
| | Other | Any other response: no response, vocalization, undefined gestures, acts of compliance (e.g. Mother says, “sit here” and points next to her, the child then walks over and sits down) |
| | Other Response | New variable created by combining the variables “gesture” and “other” for data analysis |

## Table 3

Descriptive Results of Maternal Gestures and Maternal Utterances during Mother-Child Interactions

| | Time 1 | | | Time 2 | | |
|---|---|---|---|---|---|---|
| | **M** | **SD** | **Range** | **M** | **SD** | **Range** |
| Maternal Gesture Use | 39.56 | 15.46 | 24–90 | 42.85 | 15.66 | 13–71 |
| Proximal Point | 20.26 | 14.23 | 4–63 | 24.37 | 14.77 | 5–61 |
| Any Other Gesture | 19.30 | 6.64 | 11–39 | 18.48 | 8.79 | 6–43 |
| Give/Take | 8.89 | 4.97 | 3–21 | 5.22 | 3.13 | 0–12 |
| Distal Point | 1.59 | 1.69 | 0–6 | 4.63 | 3.22 | 0–14 |
| Nod/Shrug | 2.74 | 3.77 | 0–17 | 3.33 | 2.54 | 0–9 |
| Affection | 4.37 | 4.07 | 0–14 | 1.59 | 1.28 | 0–4 |
| Maternal Utterances | | | | | | |
| Wh-Question | 5.70 | 6.15 | 0–23 | 11.93 | 10.58 | 0–36 |
| Any Other Utterance | 33.85 | 12.77 | 21–82 | 30.93 | 10.63 | 13–58 |
| Yes/No Question | 5.04 | 3.50 | 0–12 | 5.63 | 3.40 | 0–11 |
| Directive | 13.00 | 4.95 | 5–25 | 11.78 | 6.03 | 2–24 |
| Label | 8.11 | 7.04 | 1–38 | 6.04 | 3.70 | 0–17 |
| Other | 7.70 | 5.33 | 0–25 | 7.48 | 6.73 | 0–33 |

## Table 4

Frequency and Percentage of Maternal Gesture Use by Context

| | Context | | | | | |
|---|---|---|---|---|---|---|
| | **Book Reading** | **Free Play** | **Snack** | **Naturalistic 1** | **Naturalistic 2** | **Total Across Contexts** |
| Toddler Period | | | | | | |
| Frequency | 325 | 281 | 132 | 158 | 172 | 1068 |
| Percentage | 30.4% | 26.3% | 12.4% | 14.8% | 16.1% | 100.0% |
| Child Period | | | | | | |
| Frequency | 446 | 179 | 181 | 184 | 167 | 1157 |
| Percentage | 38.5% | 15.5% | 15.6% | 15.9% | 14.4% | 100.0% |
| Total Across Age Periods | | | | | | |
| Frequency | 771 | 460 | 313 | 342 | 339 | 2225 |
| Percentage | 34.7% | 20.7% | 14.1% | 15.4% | 15.2% | 100.0% |

## Table 5

Multi-Level Modeling Results for Likelihood of Child Speech Responses to Maternal Gesture Use

| **Planned Contrasts** | **Estimated Slope** | ***SE*** | ***df*** | ***t*** | ***p*** | **Odds ratio estimate** |
|---|---|---|---|---|---|---|
| Toddler Period | | | | | | |
| Mother Point vs. Other | 0.67 | 0.20 | 1063 | 3.32 | .0009 | 1.95 |
| Child Period | | | | | | |
| Mother Point vs. Other | 1.37 | 0.15 | 1152 | 9.30 | < .0001 | 3.94 |
| Combined (Toddler and Child Period) | | | | | | |
| Age period Child vs. Toddler | 1.88 | 0.12 | 2216 | 15.47 | <.0001 | 6.53 |
| Mother Point vs. Other | 1.29 | 0.12 | 2216 | 10.99 | <.0001 | 3.62 |
| Gestures Accompanying Wh-Questions | | | | | | |
| Age period Child vs. Toddler | 1.94 | 0.32 | 470 | 6.07 | < .0001 | 6.98 |
| Mother Point vs. Other | 1.60 | 0.32 | 470 | 4.94 | < .0001 | 4.93 |
| Gestures Not Accompanying Wh-Questions | | | | | | |
| Age period Child vs. Toddler | 1.70 | 0.14 | 1743 | 12.33 | < .0001 | 5.50 |
| Mother Point vs. Other | 0.82 | 0.13 | 1743 | 6.20 | < .0001 | 2.26 |
| Combined Utterances and Age periods (see Note) | | | | | | |
| Age period Child vs. Toddler | 1.75 | 0.13 | 2215 | 13.99 | < .0001 | 5.74 |
| Mother Both vs. None | 2.42 | 0.17 | 2215 | 14.37 | < .0001 | 11.24 |
| Mother Both vs. Either | 1.64 | 0.16 | 2215 | 10.31 | < .0001 | 5.14 |
| Mother Either vs. None | 0.78 | 0.13 | 2215 | 6.09 | < .0001 | 2.19 |

*Note: Both* indicates a Maternal Utterance containing *both* a point and a Wh question.

*Either* indicates a Maternal Utterance containing *either* a point and a Wh question.

*None* indicates a Maternal Utterance containing *neither* a point and a Wh question.