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Parental Responsivity and Child Communication During Mother–Child and Father–Child Interactions in Fragile X Syndrome

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ABSTRACT

Purpose: Past research shows that parentally responsive behavior toward the child positively influences language development in both neurotypical children and children with intellectual and developmental disabilities, including those with fragile X syndrome (FXS); however, most studies have focused exclusively on the mother–child relationship. The current study examined relationships between parent behavior (i.e., responsivity and behavior management) and child language performance in both mother–child and father–child interactions, as well as relationships between child characteristics and both parent behavior and child language.
Method: Participants were 23 families of young boys with FXS between 3 and 7 years of age. Mothers and fathers independently completed questionnaires assessing child characteristics and separately engaged in 12-min play-based interactions with their child via telehealth. One parent also completed a comprehensive interview assessing child adaptive behavior. Video recordings of the parent–child interactions were transcribed and coded for parent and child behavior, and measures of parent and child language were obtained from the transcripts.
Results: Mothers and fathers used similar rates of responsive behaviors during parent–child interactions, and parental responsivity was positively associated with some aspects of child language performance (i.e., talkativeness and lexical diversity). Parental behavior, however, was not associated with syntactic complexity. Older children and children with higher levels of adaptive behavior had parents who used higher rates of responsive behaviors. Fathers used higher rates of behavior management strategies compared to mothers, and this type of parent behavior was not associated with child language.
Conclusion: Overall, this study provides evidence that interventions focused on increasing parental responsiveness would be beneficial for families of children with FXS and that these interventions should be delivered early given the association between responsivity and child age.
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Individuals with fragile X syndrome (FXS) experience significant delays in multiple domains of language (Abbeduto et al., 2007). Language is an important developmental domain to understand and target for intervention in FXS given its role in a range of adaptive outcomes, including social relationships and academic success (Abbeduto & Hagerman, 1997). Children develop language through interactions with their caregivers, whose behavior changes over time to match the developmental level of the child (Brady et al., 2009); these changes in parent behavior, which are contingent on the child's developmental level and input, are considered *parental responsivity*. In recent years, research on maternal responsivity in families affected by FXS suggests that both early and sustained responsivity are

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critically important for child outcomes across a variety of domains (e.g., Brady et al., 2014, 2020; Warren et al., 2010, 2017). Moreover, recent parent-implemented language interventions (PILIs) for children with FXS between the ages of 2 and 17 years have shown that parents are able to learn and implement targeted responsive strategies (i.e., language models, recasts, open-ended questions, intonation prompts) and that there are associated gains in child engagement and language (e.g., McDuffie et al., 2018; McDuffie, Machalicek, et al., 2016; McDuffie, Oakes, et al., 2016; Thurman et al., 2020). However, most past research on the role of responsive parenting in FXS, and on PILIs, has focused exclusively on the mother–child dyad. The aims of the current study were (a) to examine relationships among maternal responsivity, paternal responsivity, and child language performance in parent–child dyadic interactions, as well as the relationships between child characteristics and child language performance, and (b) to examine relationships between child characteristics and both maternal and paternal responsivity. In this study, we expanded the focus beyond the mother–child dyad to also include examinations of these relationships in father–child dyads.

Behavioral Phenotype of FMR1-Associated Conditions

FXS, an X-linked disorder, is the leading inherited cause of intellectual disability (ID; Crawford et al., 2001). FXS results from the expansion of a cytosine–guanine–guanine (CGG) trinucleotide sequence in the *FMR1* gene to greater than 200 repeats, which is defined as the full mutation (Oostra & Willemsen, 2003). Because FXS is X-linked, males tend to be affected more often and more severely than females. Specifically, it is estimated that approximately one in 7,143 males are affected by the full mutation compared to only approximately one in 11,111 females (Hunter et al., 2014). Additionally, most males with FXS have IQ scores in the range of ID (i.e., typically < 70), whereas only 25%–50% of females with FXS have scores that meet criteria for ID (Hessl et al., 2009; Wright-Talamante et al., 1996). Due to these sex-based differences, the current study focused only on males with FXS.

Other phenotypic characteristics of FXS, all of which are likely to interfere with language learning and social interaction, include hyperactivity (Baumgardner et al., 1995), limitations in executive functioning (Loesch et al., 2003), anxiety and social withdrawal (Cordeiro et al., 2011), and aggression (Hessl et al., 2008). Males with FXS also frequently display characteristics of autism spectrum disorder (ASD), with as many as 50%–60% of males receiving a diagnosis of ASD (Abbeduto et al., 2019). Moreover, individuals with FXS seem to experience more significant impairment in social functioning compared to individuals with other genetic neurodevelopmental disorders, perhaps in part because of the increased rates of hyperactivity, impulsivity, and inattention observed in FXS (Chromik et al., 2019).

Only females can transmit the *FMR1* full mutation to their biological children. Biological mothers of children with FXS are most often *FMR1* premutation carriers, although some may have the full mutation. *FMR1* premutation carriers have between 55 and 200 CGG repeats and present with a unique phenotype that is shaped by genetic and environmental factors (Mailick et al., 2018; Seltzer et al., 2012). The *FMR1* premutation can result in two established disorders: fragile X–associated primary ovarian insufficiency and fragile X–associated tremor ataxia syndrome, a late-onset neurogenerative disease that affects both male and female premutation carriers (Visootsak et al., 2014). Females with the *FMR1* premutation are also at risk by virtue of their biology for a range of psychiatric, medical, and cognitive differences compared to the general population (Hagerman et al., 2018). For example, female premutation carriers are more likely to experience mood or anxiety disorders compared to the general population (Bourgeois et al., 2011). These women are also more likely to experience medical problems, including migraines, fibromyalgia, neuropathy, and vestibular difficulties compared to females without the premutation, as well as problems in executive functioning, attention, working memory, arithmetic, and some aspects of language (Wheeler et al., 2014). Mothers of children with FXS are also likely to experience high levels of parenting stress (Hartley et al., 2012) and mental health challenges (Abbeduto et al., 2004), due at least in part to the significant challenges associated with parenting a child with high levels of challenging behaviors, low levels of adaptive behaviors, and the associated increased financial burden and social isolation (Minnes et al., 2015; Tint & Weiss, 2016).

The cumulative effects of the factors affecting females with the *FMR1* premutation or full mutation may constrain the development of a warm and responsive mother–child relationship with subsequent negative impacts on child development (Lovejoy et al., 2000; Warren & Brady, 2007). These challenges faced by families affected by FXS are also likely to contribute to reduced marital satisfaction and family cohesion (Baker et al., 2012), which could further negatively affect both the mother–child and father–child relationships and thereby the child's development across multiple domains. Although not all families of children with FXS experience suboptimal parent–child or mother–father relationships, these relationships are important to investigate given the elevated levels of mental health challenges and parenting stress experienced by both mothers and fathers of children with FXS (McCarthy et al., 2006; Potter, Harvey, et al., 2022).

Theoretical Framework

Both the transactional model (e.g., Roberts et al., 2014; Sameroff & Chandler, 1975; Woynaroski et al., 2014) and family systems theory (e.g., Bornstein & Sawyer, 2006; Cox & Paley, 1997; Trivette et al., 2010) explain how responsive parenting can influence child development across multiple domains, including language. These models also provide an explanatory framework for the social interactionist approach to language learning (e.g., Brady et al., 2009; Chapman, 2000; Warren & Brady, 2007). Family systems theory focuses on the importance of the family as an ecological system in which an individual develops such that a person cannot be fully understood without considering the context of the family in which they were raised (Cox & Paley, 1997). The transactional model suggests that the development of a child results from the bidirectional effects between the child and the environment, such that experiences in the environment are not considered independent of the child. From birth onward, a child's relationship with their parents affects socioemotional, behavioral, and cognitive outcomes. The bidirectional parent–child relationship suggests that "parent behaviors are both the cause and the consequence of child behaviors" (Laursen & Collins, 2009, p. 11). Features of the parent, including parental physical and mental health, parenting practices, and parental perceptions, as well as features of the child, including cognitive level, temperament, and the ability to self-regulate, make for transactional interactions in which the parent and child are an interdependent unit (e.g., Belsky, 1984; Neece et al., 2012; Sameroff, 2009; Sameroff & Mackenzie, 2003). Thus, early-onset conditions (e.g., genetic disorders) lead to later outcomes (e.g., impairments vs. growth in language) through bidirectional transactions between the child and their environment (e.g., interactions with a parent).

Language Learning and Responsive Parenting

Children learn language by engaging in back-and-forth interactions with more advanced communicative partners, such as their parents or other adult caregivers (e.g., Bruner, 1975; Ford et al., 2020; Golinkoff et al., 2018; Sameroff & Fiese, 2000). According to the social interactionist approach to language development, as children become more advanced communicators, adults respond by adjusting their behaviors to match the child's developmental level (Warren & Brady, 2007). These modifications in reaction to the child's developmental level are examples of responsivity (Brady et al., 2009). For example, mothers often use a slower rate of speech, exaggerated prosody, and more simplified language when talking to infants and very young children compared to their talk to older children and adults (i.e., infant/child-directed speech; Ma et al., 2011; Newman et al., 2016).

As children become more communicative and socially engaged, responsive parents modify their behaviors and adapt to their child's developing abilities and interests. Examples include maintaining their child's focus of attention and following the lead of their child through behaviors such as commenting and recasting their child's activities and interests (Brady et al., 2009; Tamis-LeMonda et al., 2014). In typical development, the degree of maternal responsiveness has been found to be predictive of the timing of early language milestones, including first imitations, first words, attainment of first 50 expressive words, first combinations, and first use of language to talk about the past (Tamis-LeMonda et al., 2001). Moreover, consistent, or sustained, responsiveness over time has been shown to be important for cognitive and social development throughout early childhood (Landry et al., 2001).

Parental responsivity has also been found to have positive effects on various developmental domains—including language development—in populations with neurodevelopmental disabilities. For example, Yoder and Warren (1998) found that parental responsivity was predictive of the display of intentional communication by young children with developmental disabilities of various etiologies. Maternal responsivity was also found to have a positive influence on the relationship between children's intentional communication and later language development (Yoder & Warren, 1999), as well as an effect on children's receptive and expressive language 6 and 12 months after participation in two different prelinguistic communication interventions (Yoder & Warren, 2001). Moreover, McDuffie and Yoder (2010) found that certain behaviors reflective of parental verbal responsiveness predicted early vocabulary acquisition in young children with ASD. In another study, Sterling and Warren (2014) found that mothers of children with Down syndrome (DS) were able to employ a highly responsive and interactive style of parenting that was facilitative of their child's linguistic development, particularly for children in the sample who were older and more communicative. That is, the more mothers adapt to their children's linguistic growth by increasing their own use of facilitative behaviors that match their child's current level of functioning and need, the more positive the children's outcomes.

Maternal Responsivity and Child Outcomes in FXS

Over the past decade, Warren, Brady, Sterling, and colleagues have investigated longitudinal relationships between maternal responsivity and child outcomes in a sample of 55 mother–child dyads. Warren et al. (2010) examined the effects of maternal responsivity (i.e., gesture use, requests for verbal compliance, comments, and recodes) on language development across 3 years in young children with FXS. They found that maternal responsivity predicted both proximal and distal levels of receptive and expressive language at 36 months, even after controlling for children's ASD symptoms and nonverbal developmental level. Similarly, Brady et al. (2014) found that sustained responsivity measured across 4 years predicted later receptive and expressive vocabulary development up to 9 years of age. In a more recent study, Brady et al. (2020) found that maternal responsivity continued to be important for language development during adolescence in this sample. Specifically, maternal commenting (i.e., any comments that maintain the child's focus of attention, relate to the child's actions/interests at the time, or are in response to something the child is doing or saying) was related to growth in child rate of different words produced in conversation samples as well as receptive vocabulary as measured by a standardized test. However, maternal commenting was not related to growth in either expressive vocabulary (as measured by a standardized test) or expressive syntax, which was consistent with previous findings in this sample (Komesidou et al., 2017). These findings suggest not only that maternal responsivity in FXS is important in early childhood (Warren et al., 2010) and middle childhood (Brady et al., 2014) but also that sustaining responsivity has a positive influence on some aspects of language development even throughout adolescence. Therefore, responsivity could be a potential target for intervention even beyond the early years of development for this population.

The effects of maternal responsivity in FXS extend beyond language development. In another study of their mother–child dyads, Warren et al. (2017) examined the relationship between maternal responsivity and adaptive behavior as measured by the Vineland Adaptive Behavior Scales (Sparrow et al., 1984, 2005). Overall, they found that sustained maternal responsivity had a significant and positive impact on growth in child Communication domain scores, even after controlling for symptoms of ASD and developmental level. In addition, maternal responsivity predicted trajectories of skills in the Socialization and Daily Living Skills domains, but to a lesser extent than for the Communication domain. Perhaps the most interesting finding from this study was that roughly half of the children showed declines in adaptive behavior (i.e., decreases in raw scores over time); yet, those participants who had mothers who were more responsive declined less than those who had mothers who were less responsive. This finding was most evident for the Communication domain, suggesting the importance of responsive parenting for the development of adaptive communication skills during middle childhood. The results of these studies highlight the importance of maternal responsivity for child language and adaptive functioning outcomes in FXS.

The Role of Fathers in Child Development

Little is known about the ways in which fathers of children with FXS influence the child's development or the impact of the child on the father, including how the child affects the father's well-being and behavior (Riley et al., 2017). Past research has shown that high-quality paternal involvement is associated with improved outcomes for neurotypical children above and beyond the outcomes associated with high-quality maternal involvement (Flippin & Watson, 2015). High levels of father involvement have also been shown to be positively associated with marital satisfaction, parental competence, and closeness to neurotypical children in parents (Ehrenberg et al., 2001). In contrast, low levels of paternal involvement, potentially caused by stress or depression, are related to psychological and emotional dysfunction in neurotypical children, as well as decreased rates of cognitive and language development (Kane & Garber, 2004; Paulson et al., 2009; Wanless et al., 2008). Evidence also suggests that father–child interactions support the neurotypical child's ability to regulate their emotions and arousal (Bocknek et al., 2017; Feldman, 2003). The father's role in shaping child outcomes warrants further attention in families of children with FXS.

Unfortunately, more often than not, the father's role in the family has been ignored from both an empirical and a societal standpoint, leading to an exclusive focus and, as a result, an increased burden on the mother such that her role, either positive or negative, in influencing child outcomes is more likely to be overstated (Wilson & Prior, 2011). Understanding more about the role of fathers in shaping the development of children with FXS is important for several reasons, including fathers' increasing role in caregiving responsibilities in recent decades and the benefit of having both mothers and fathers involved in the child's therapies and interventions (Fox et al., 2015; Wang et al., 2006). Father involvement in the child's interventions, including parent-implemented interventions that target paternal behavior, could lead to increased parental competence and decreased stress, as well as improved coparenting and higher mother–father relationship quality (Bronte-Tinkew et al., 2007; Flippin & Watson, 2015). The current study was designed to replicate some of the past findings regarding maternal responsivity in a new sample of mother–child dyads and to extend the investigation to also include father–child dyads.

Current Study

The first aim of the current study was to examine relationships among maternal responsivity, paternal responsivity, and child language in parent–child dyadic interactions, as well as relationships between child characteristics (i.e., challenging behaviors, ASD characteristics, and adaptive behavior) and child language. We hypothesized that higher rates of maternal and paternal responsivity in the dyadic interactions would be positively associated with better child language performance (e.g., Warren & Brady, 2007; Warren et al., 2010). The second aim was to examine relationships between child characteristics and both maternal and paternal responsivity. We hypothesized that higher levels of child adaptive behavior as well as fewer challenging behaviors and characteristics of ASD would be associated with higher rates of parental responsivity (Brady et al., 2014; Fielding-Gebhardt et al., 2023; Warren et al., 2010).

Method

Participants

Participants were 23 families of male children with FXS between the ages of 3.00 and 7.99 years, with a total of 69 participants: 23 fathers (22 biological fathers and one stepfather), 23 biological mothers, and 23 male children with FXS. Only families of boys with FXS were included in the study because nearly all males with FXS have ID and language impairment, whereas females with FXS demonstrate more variability in their intellectual functioning and abilities (Abbeduto et al., 2007; Hessl et al., 2009). To be eligible to participate in the study, the child had to be living at home with both parents and have no uncorrected sensory or motor impairments that would limit his ability to participate in the study. Additionally, English had to be the primary language spoken in the home. Parents were asked to provide documentation of their child's diagnosis of FXS as well as the mother's *FMR1* premutation or full mutation status if available. Medical reports were required to confirm the child's diagnosis of the *FMR1* full mutation, but verbal confirmation was accepted for the mother's genetic status. The study was approved by the institutional review board at the University of California (UC), Davis, in advance of recruitment, and both parents provided informed consent electronically via REDCap (Research Electronic Data Capture; Harris et al., 2009, 2019).

Family demographic characteristics are presented in Table 1. Over 80% of participants identified as White and not Hispanic or Latine. Approximately 74% of mothers and 70% fathers in the study had a bachelor's degree, master's degree, or other advanced degree, and annual household income was $100,000 or above for more than half of the families in the study. All families resided in North America, with families living in 13 U.S. states and two Canadian provinces. This sample of participants and some of these data have been previously described in Potter, Harvey, et al. (2022), which focused on features of, and relationships between, parental well-being, couple well-being, and child characteristics.

Procedure and Measures

The data for the current study were collected as part of a larger study investigating relationships within families and parent and child behavior in families affected by FXS. Participation in the study involved multiple calls with an examiner via secure distance teleconferencing (i.e., Skype for Business or Zoom), the completion of online questionnaires, and an interview. Data were collected between December 2019 and July 2021. Therefore, most families were tested during the COVID-19 pandemic, with only two families completing their participation in the study prior to the first community-diagnosed case in California on February 23, 2020.

Child Measures

Mothers and fathers independently completed two questionnaires to assess child characteristics: the Aberrant Behavior Checklist–Second Edition (ABC-2; Aman & Singh, 2017) and the Social Responsiveness Scale–Second Edition (SRS-2; Constantino & Gruber, 2012). One parent also completed the Vineland Adaptive Behavior Scales–Third Edition (Vineland-3; Sparrow et al., 2016) as an interview to assess the child's adaptive behavior.

The ABC-2 and SRS-2 were administered as online surveys via REDCap, a secure web-based software platform designed to support data capture for research studies, providing (a) an intuitive interface for validated data capture, (b) audit trails for tracking data manipulation and export procedures, (c) automated export procedures for seamless data downloads to common statistical packages, and (d) procedures for data integration and interoperability with external sources.

The ABC-2 is a 58-item scale that measures challenging behaviors of individuals with developmental disabilities in six domains: Irritability, Socially Unresponsive/Lethargic, Stereotypy, Hyperactivity, Inappropriate Speech, and Social Avoidance. Total raw scores from the FXS-specific subscale scoring (Sansone et al., 2012) were used in analyses; three items were omitted in the exploratory factor analysis in Sansone et al. (2012), and thus total raw scores range from 0 to 165, with higher scores

Table 1. Family demographic characteristics.

Individual characteristics	Child	Mother	Father
Age (years)			
M (*SD*)	5.68 (1.45)	38.28 (6.00)	40.16 (5.86)
Range	3.07–7.90	25.15–50.43	27.79–51.46
Race: *n* (%)			
White	20 (87)	21 (91)	20 (87)
Asian	2 (9)	2 (9)	3 (13)
Mixed/Multiracial	1 (4)	0 (0)	0 (0)
Ethnicity: *n* (%)			
Not Hispanic/Latine	20 (87)	20 (87)	19 (83)
Hispanic/Latine	3 (13)	3 (13)	4 (17)
Parent characteristics		**Mother**	**Father**
Education: *n* (%)			
Some high school		0 (0)	1 (4)
High school/GED		1 (4)	2 (9)
Some college/technical school		3 (13)	2 (9)
Associate's/technical degree		2 (9)	2 (9)
Bachelor's degree		8 (35)	9 (39)
Master's/other advanced degree		9 (39)	7 (30)
Employment: *n* (%)			
Not currently employed		9 (39)	4 (17)
Part-time		7 (30)	0 (0)
Full-time		7 (30)	19 (83)
Family characteristics			
Annual household income: *n* (%)			
Under $50,000	1 (4)		
$50,001–$100,000	8 (35)		
$100,001–$150,000	5 (22)		
$150,001–$250,000	7 (30)		
Unknown	2 (9)		

Note. The individual percentage values are rounded and may not total 100%. GED = General Educational Development.

reflecting more severe challenging behavior. This scale takes approximately 10 min to complete.

The SRS-2, a 65-item scale, measures social impairments commonly associated with ASD, providing DSM-5–compatible subscale scores for Social Communication and Interaction and Restricted Interests and Repetitive Behavior, as well as a Total *T* score. Mothers and fathers independently completed either the preschool (2½–4½ years) or school-aged (4–18 years) form depending on their child's chronological age. Total *T* scores from the SRS-2 were used in analyses. The scale takes approximately 15 min to complete.

The Vineland-3 measures adaptive behavior across multiple domains. For the current study, the Adaptive Behavior Composite score as well as the Communication, Daily Living Skills, and Socialization domain standard scores were used in analyses. The Vineland-3 was administered by a trained examiner using Q-Global, a web-based platform for online administration. The child's primary caregiver (as reported by the parents) participated in the interview via the phone or a secure teleconferencing platform. The Vineland-3 interview takes approximately 1–2 hr to complete.

Dyadic Interactions

Each family who participated in the study was loaned a set of developmentally appropriate toys, including a puzzle, DUPLO blocks, a garbage truck, a farm set, and a pretend breakfast food set. On different days of the study, mothers and fathers were instructed to play with their child as they usually would for 12 min. Families were told that they could also include toys of their own in the play interaction if they desired. The play interactions were recorded using secure teleconferencing. During the play interaction, the examiner turned off her camera and muted her microphone. Immediately after the sample, the examiner asked the parent whether the child's behavior during the interaction was typical in comparison to their usual interactions to ensure that a representative sample was collected.

Transcription

Video recordings of the dyadic play-based interactions were transcribed by trained research assistants using Systematic Analysis of Language Transcripts (SALT; Miller & Iglesias, 2008). SALT is a software program that standardizes the process of transcribing and analyzing language samples. The dyadic samples were transcribed according to the procedures described in Abbeduto et al. (2020). In these procedures, a primary transcriber completes a first draft of a transcript, which is then reviewed and edited by a second transcriber. The primary transcriber then finalizes the transcript based on the feedback from the second transcriber. These procedures consistently yield an average interrater transcript reliability of approximately 90% or above in language samples of participants with FXS (Abbeduto et al., 2020; Kover et al., 2012; Nelson et al., 2018).

Measures of parent and child language can be automatically generated from finalized SALT transcripts, including total number of complete and intelligible (C&I) utterances or conversational turns (i.e., a measure of talkativeness), number of different words (NDW; a measure of lexical diversity), and mean length of utterance in morphemes (MLUm; a measure of syntactic complexity). These outcomes were generated separately for the mother–child and father–child interactions.

Coding of Parental Behavior

After the video recordings of the dyadic interactions were transcribed, the transcripts were coded while viewing the videos for the presence of various parental behaviors utilizing a coding scheme adapted from Warren and colleagues (e.g., Sterling et al., 2013; Warren et al., 2010). When the parent had multiple utterances in succession, only the final utterance prior to either a 3-s pause or a child communication act was coded. See Table 2 for definitions and examples of the parental behavior codes. Additionally, the parent–child interaction coding manual is provided in Supplemental Material S1. Composite scores for parental responsivity and behavior management were based on frequency counts of the observed behaviors within each category. Proportion scores for these variables were also calculated as the total composite score in each category divided by the total number of C&I parent utterances in the transcript. For example, a parent who had 144 responsive utterances, 17 behavior management utterances, and 227 total C&I utterances would have a responsivity proportion of 144/227 = 0.63 and a behavior management proportion of 17/227 = 0.07.

Table 2. Parental behavior codes and definitions.

Category and behavior	Definition	Examples
Parental responsivity		
Comments	All comments that maintain the child's focus of attention, relate to the child's actions and interests at the time, or are in response to something the child is doing or saying	Talking about what the child or parent can see, hear, smell, taste, or touch: "That's bumpy." Praise in reaction to something the child has done: "Good job!"
Requests for verbal compliance	Parent questions or statements intended to elicit a verbal response from the child that relate to the child's focus of attention	All questions that require a verbal response from the child: "What color is the truck?" Parent asks the child to say something: "Can you say, 'choo-choo?'"
Recodes	Verbal interpretation of the child's communication act that extends the form of the child's utterance	Parent reproduces a content word in a reasonable interpretation of the child's verbal act: Child says, "Ball." Parent says, "That's a big blue ball!"
Behavior management		
Request for behavioral compliance	Parent questions or statements intended to elicit a behavioral response from the child	Look/See statements that are followed by a directive: "Look, put it on top like this." Let/Let's statements intended to get child to do something: "Let's put these animals in the barn."
Redirects	The parent directs the child to engage with something that is outside of the child's current focus of attention	The child is playing with a toy, and the parent instructs or asks the child to do something different: "Let's put away the food and do the puzzle now."
Zaps	Parent directives that limit, restrict, or discipline the child's behavior in some way	Examples of verbal restrictions: "Be careful!" "Don't do that!"

Coding reliability. Four undergraduate research assistants were trained through group consensus coding to utilize the adapted coding scheme. Each transcript was independently coded by two research assistants. Following independent coding, transcripts were compared, and disagreements were resolved via consensus coding. Interobserver agreement of total scores for parental responsivity and behavior management codes was based on a random sampling of approximately 20% of the sessions. We compared independently coded (i.e., preconsensus) transcripts. Two-way random intraclass correlation coefficients (ICCs) were .994 for the parental responsivity composite and .937 for the behavior management composite. Additionally, ICCs for the subcategories ranged from .809 to .996. The only categories with ICCs below .850 were low-frequency parent behaviors (i.e., recodes, redirects, and zaps), all of which had mean frequencies below 2.

Analysis Plan

All variables were visually inspected to check for model assumptions of normality. Tests for skewness and kurtosis were also examined. Transformations and nonparametric alternatives were considered for any data that did not meet parametric assumptions. Descriptive summaries the measures of child characteristics and the measures of the parent behavior and parent and child language measures from the dyadic interactions were reported. Interspousal correlations were calculated to determine the degree of correspondence between parent behavior and parent and child language measures in the mother–child and father–child interactions. Comparisons of means for parent behavior and parent and child language measures from the mother–child and father–child interactions were also reported using paired-samples *t* tests and Wilcoxon signed-ranks tests. Then, correlations between the dependent and independent variables for the study aims were reported to investigate relationships between the variables, examine potential differences between these relationships for mothers and fathers, and assess for multicollinearity between the independent variables for Aims 1 and 2. Correlations between (a) maternal ratings of the child and mother–child interaction variables, (b) paternal ratings of the child and father–child interaction variables, and (c) combined parental ratings of the child and parent–child interaction variables were reported. The combined parental ratings were based on means of the variables (as opposed to sums of the variables within families).

The dependent variables for Aim 1 were the following child language measures obtained from the SALT transcripts of the dyadic play-based language samples: total number of C&I utterances, NDW, and MLUm. The independent variables for Aim 1 were proportion scores for parental responsivity and behavior management from the mother–child and father–child dyadic interactions, as well as child characteristics embodied in the ABC-2 Total raw scores, SRS-2 Total T-scores, the Vineland-3 domain and Adaptive Behavior Composite scores, and child age. For Aim 2, the primary outcome variables were proportion scores for parental responsivity and behavior management from the mother–child and father–child dyadic interactions and the independent variables were child characteristics (i.e., those listed above for Aim 1). For Aims 1 and 2, models were specified using a multilevel modeling (MLM) approach (Raudenbush & Byrk, 2002), given the nonindependence of the data collected from different parent–child dyads within families. In this approach, the data from each dyad are nested within a group that has an *N* of 2 (Campbell & Kashy, 2002). Effect coding was used for parent sex (i.e., male = 1 and female = −1), and continuous predictors were centered to their respective grand means.

For Aim 1, ICCs were calculated to estimate the proportion of the total variation in the child language measures (i.e., total number of C&I utterances, NDW, and MLUm) that exists between versus within families. Then, separate models for child total number of C&I utterances, child NDW, and child MLUm were specified to investigate the contributions of parent behavior and child characteristics to child language performance. There were strong and significant correlations between the variables for child ASD characteristics, child challenging behavior, and child adaptive behavior; however, there was not a significant correlation between the measures of child challenging behavior and child adaptive behavior. Therefore, in addition to child age, the ABC-2 Total raw score and the Vineland-3 Adaptive Behavior Composite score were used in the models for Aim 1 to predict child language performance. Parent sex was also included as a predictor in the models for Aim 1 to examine whether there were differences in the child language performance between the mother–child and father–child interactions.

For Aim 2, ICCs were calculated to estimate the proportion of the total variation in the measures of parental responsivity and behavior management that exists between versus within couples. Then MLMs were specified to investigate the contributions of child characteristics to parental responsivity and behavior management. The same child variables used in Aim 1 were used in Aim 2 to predict parent behavior. Parent sex was also included as a predictor.

Results

Aim 1 was designed to examine the relationships among maternal responsivity, paternal responsivity, and child language performance in parent–child dyadic interactions, as well as the relationships between child characteristics and child language performance. Table 3 displays descriptive statistics for the measures of child characteristics (i.e., scores on the ABC-2, SRS-2, and Vineland-3). Aim 2 was designed to examine relationships between child characteristics and parental responsivity. Table 4 displays descriptive statistics for the parent behavior variables as well as the parent and child language variables that were coded or obtained from the transcripts of the mother–child and father–child dyadic play-based interactions. Paired-samples *t* tests and Wilcoxon signed-ranks tests (when appropriate) confirmed that there were no statistically significant differences between child language performance in the mother–child and father–child interactions, mothers' and fathers' language use, or mothers' and fathers' use of responsivity or behavior management strategies, with the exception of parent talkativeness, that is, total number of C&I utterances, $t(22) = 3.22$, $p = .004$; parent lexical diversity, that is, NDW, $t(22) = 4.29$, $p < .001$; and the behavior management proportion score, $Z = -2.01$, $p = .031$. Mothers had a greater number of total C&I utterances and NDW compared to fathers, and fathers had a higher proportion score for behavior management strategies compared to mothers.

Table 4 also displays interspousal correlations; given that some of these variables were not normally distributed, Spearman's rank-order correlations were reported instead of Pearson's correlations. Interspousal correlations indicated that there were significant correspondences between the measures of mothers' and fathers' language as well as their responsivity frequency and proportion totals. There were also significant correspondences between the measures of child language in the mother–child and father–child interactions. No significant correspondences were

Table 3. Measures of child characteristics.

Measure	*M* (*SD*) Range: Mothers	*M* (*SD*) Range: Fathers
ABC-2 domain		
Irritability	18.26 (10.62) 1–38	17.44 (11.54) 1–47
Socially unresponsive/lethargic	4.52 (5.86) 0–23	5.17 (4.65) 0–20
Stereotypy	6.09 (2.78) 1–12	4.96 (3.76) 0–13
Hyperactivity	14.04 (8.07) 2–27	11.00 (5.42) 1–24
Inappropriate speech	4.48 (3.85) 0–11	3.17 (2.29) 0–8
Social avoidance	1.39 (2.61) 0–9	1.52 (2.39) 0–9
Total score	49.39 (27.71) 12–98	43.87 (24.70) 10–103
SRS-2 domain		
Social communication and interaction	69.00 (11.00) 49–90	66.83 (9.53) 49–85
Restricted interests and repetitive behavior	75.48 (11.63) 57–92	69.17 (10.35) 54–90
Total score	70.87 (10.96) 51–91	67.57 (9.40) 51–87
Vineland-3 domain		
Communication	62.65 (18.31) 20–96	
Daily living skills	69.13 (10.09) 52–93	
Socialization	72.52 (14.51) 40–104	
Adaptive behavior composite	67.70 (11.87) 39–89	

Note. ABC-2 = Aberrant Behavior Checklist–Second Edition; SRS-2 = Social Responsiveness Scale–Second Edition.

Table 4. Comparisons of parental behavior and child language during dyadic parent–child interactions.

Language	*M* (*SD*) Range Mother–child interaction	 Father–child interaction	*ρ* (*p* value) Interspousal correlation
Parent language			
Total C&I utterances	288.52 (62.22) 150–405	240.30 (82.97) 85–450	**0.49*** **(.017)**
Number of different words	211.48 (46.71) 126–307	171.57 (49.46) 74–271	**0.55**** **(.007)**
Mean length of utterance – morphemes	3.36 (0.51) 2.21–4.53	3.41 (0.50) 2.43–4.25	**0.42*** **(.047)**
Total parental responsivity (frequency)	93.39 (42.91) 26–176	82.48 (39.58) 11–149	**0.81***** **(< .001)**
Total parental responsivity (proportion of total C&I utterances)	0.33 (0.14) 0.07–0.61	0.35 (0.15) 0.13–0.60	**0.69***** **(< .001)**
Total behavior management (frequency)	14.43 (12.41) 5–53	17.57 (12.78) 4–58	0.34 (.116)
Total behavior management (proportion of total C&I utterances)	0.05 (0.04) 0.01–0.14	0.07 (0.04) 0.02–0.19	0.29 (.177)
Child language			
Total C&I utterances	132.17 (87.29) 6–307	138.13 (89.34) 1–303	0.81*** **(< .001)**
Number of different words	68.13 (48.04) 1–154	71.09 (55.61) 1–178	**0.88***** **(< .001)**
Mean length of utterance – morphemes	1.70 (0.55) 1.00–2.75	1.82 (0.77) 1.00–3.88	**0.90***** **(< .001)**

Note. C&I = complete and intelligible.
*$p < .05$. **$p < .01$. ***$p < .001$.

found between mothers' and fathers' use of the individual behavior management strategies or the total frequency or proportion scores for behavior management.

Tables 5 and 6 display correlations between measures of parent behavior, child language, and child characteristics for mothers and fathers, respectively, and Table 7 displays correlations between these variables combined across parents. Table 5 shows that there were significant positive correlations between maternal responsivity and the following variables: child age, child total C&I utterances, child NDW, child MLUm, and the Vineland-3 Daily Living Skills and Socialization domain scores and the Vineland-3 Adaptive Behavior Composite score. There were no significant correlations between maternal behavior management and child language or child characteristics apart from a significant correlation between maternal behavior management and the Vineland-3 Socialization domain score. Table 6 shows that there were significant correlations between paternal responsivity and the following variables: child total C&I utterances, child NDW, child MLUm, and child ASD characteristics, as well as all Vineland-3 domain scores and the Vineland-3 Adaptive Behavior Composite score. Additionally, there were no significant correlations between paternal behavior management and any of the measures of child language or child characteristics.

Table 7 shows that when the maternal and paternal variables are analyzed together, parental responsivity was associated with child age, ASD characteristics, adaptive behavior, and all child language measures, and parental behavior management was only associated with the Vineland-3 Socialization domain. Child age was associated with all child language measures, but not with child challenging behaviors, ASD characteristics, or adaptive behavior. All child language measures were associated with adaptive behavior, and child lexical diversity and syntactic complexity were associated with ASD characteristics.

Aim 1 Models: Influence of Parent Behavior on Child Language

The ICC for child talkativeness indicated that 88.1% of the variation was due to between-dyad factors, whereas 11.9% was due to within-dyad factors. For child lexical diversity, 93.2% of the variation was due to between-dyad factors, whereas 6.8% was due to within-dyad factors. For child syntactic complexity, 81.1% of the variation was due to between-dyad factors, whereas 18.9% was due to within-dyad factors. Table 8 presents the results of the MLM analyses for Aim 1. The lexical diversity and syntactic complexity variables were square root–transformed to reduce positive skewness.

Table 5. Spearman correlations for maternal ratings of the child and mother–child interaction variables (n = 23).

Variable	1	2	3	4	5	6	7	8
1. Responsivity	1.00							
2. Behavior management	−0.16 (.471)	1.00						
3. Child age	**0.42* (.050)**	0.06 (.799)	1.00					
4. Child total C&I utterances	**0.81*** (< .001)**	0.18 (.399)	**0.47* (.025)**	1.00				
5. Child NDW	**0.84*** (< .001)**	−0.01 (.993)	**0.47* (.023)**	**0.94*** (< .001)**	1.00			
6. Child MLUm	**0.68*** (< .001)**	−0.10 (.666)	**0.47* (.025)**	**0.75*** (< .001)**	**0.89*** (< .001)**	1.00		
7. Challenging behavior[a]	−0.03~ (.089)	0.37~ (.080)	0.09 (.687)	0.23 (.290)	0.16 (.468)	0.19 (.393)	1.00	
8. ASD characteristics[b]	−0.27 (.209)	0.40~ (.059)	0.36~ (.093)	−0.05 (.805)	−0.20 (.369)	−0.17 (.450)	**0.47* (.023)**	1.00
9. Communication[c]	0.41~ (.054)	−0.21 (.343)	−0.25 (.257)	0.39~ (.070)	**0.44* (.036)**	**0.45* (.031)**	−0.10 (.653)	**−0.63** (.001)**
10. Daily living skills[d]	**0.43* (.043)**	−0.27 (.209)	−0.17 (.438)	**0.45* (.031)**	**0.52* (.011)**	0.40~ (.059)	−0.24 (.273)	**−0.65*** (< .001)**
11. Socialization[e]	**0.48* (.021)**	**−0.43* (.042)**	−0.19 (.378)	0.34 (.118)	**0.43* (.041)**	0.39~ (.066)	−0.36~ (.094)	**−0.83*** (< .001)**
12. Vineland-3 ABC[f]	**0.46* (.026)**	−0.21 (.326)	−0.25 (.259)	**0.45* (.032)**	**0.50* (.015)**	**0.43* (.039)**	−0.22 (.315)	**−0.78*** (< .001)**

Note. The bold values indicates significant values. Proportion scores used for responsivity and behavior management variables. C&I = complete and intelligible; NDW = number of different words; MLUm = mean length of utterance in morphemes; ASD = autism spectrum disorder.

[a]ABC-2 total raw score. [b]SRS-2 total T score. [c]Vineland-3 Communication domain standard score. [d]Vineland-3 Daily Living Skills domain standard score. [e]Vineland-3 Socialization domain standard score. [f]Vineland-3 Adaptive Behavior Composite score. ~$p < .10$.

*$p < .05$. **$p < .01$. ***$p < .001$.

Prediction of Child Talkativeness: Total Number of C&I Utterances

As expected, there was a significant main effect of parental responsivity on child talkativeness ($p < .001$). Across all dyads, when the other predictors were held constant, children whose parents displayed greater-than-average rates of responsive behaviors were more talkative compared to children whose parents displayed less-than-average rates of responsive behaviors. There was also a significant main effect of adaptive behavior ($p = .010$). Across all dyads, when the other predictors were held constant, children with above-average levels of adaptive behavior were more talkative compared to children with below-average levels of adaptive behavior. Additionally, there was a main effect of child age ($p < .010$), suggesting that across all dyads, when the other predictors were held constant, older children were more talkative than younger children. There were no significant main effects of either challenging behavior or parent sex on child talkativeness.

Prediction of Child Lexical Diversity: NDW

As with child talkativeness, there were significant main effects of parental responsivity ($p < .001$), child adaptive behavior ($p < .001$), and child age ($p = .001$) on child lexical diversity, but no significant main effects of either challenging behavior or parent sex on child lexical diversity. Specifically, across all dyads, when the other predictors were held constant, children whose parents displayed greater-than-average rates of responsive behaviors had higher levels of lexical diversity compared to children whose parents displayed lower-than-average rates of responsive behaviors. Additionally, across all dyads, when the other predictors were held constant, children with above-average levels of adaptive behavior had higher levels of lexical diversity compared to children with below-average levels of adaptive behavior. Finally, across all dyads, when the other predictors were held constant, older children had higher levels of lexical diversity than younger children.

Prediction of Child Syntactic Complexity: MLUm

Unlike the other child language measures, there was not a significant main effect of parental responsivity on syntactic complexity ($p = .159$). There were, however, significant main effects of both child adaptive behavior ($p = .001$) and child age ($p < .001$) on syntactic complexity. Across all dyads, when the other predictors were held constant, children with above-average levels of adaptive behavior had higher levels of syntactic complexity compared to

Table 6. Spearman correlations for paternal ratings of the child and father–child interaction variables (n = 23).

Variable	1	2	3	4	5	6	7	8
1. Responsivity	1.00							
2. Behavior management	–0.25 (.250)	1.00						
3. Child age	0.27 (.213)	0.29 (.173)	1.00					
4. Child total C&I utterances	**0.83*** (< .001)**	0.05 (.809)	0.33 (.130)	1.00				
5. Child NDW	**0.81*** (< .001)**	0.04 (.849)	0.40~ (.061)	**0.94*** (< .001)**	1.00			
6. Child MLUm	**0.76*** (< .001)**	–0.07 (.757)	**0.46* (.025)**	**0.85*** (< .001)**	**0.93*** (< .001)**	1.00		
7. Challenging behavior[a]	–0.32 (.136)	–0.20 (.356)	–0.28 (.198)	**–0.47* (.024)**	**–0.46* (.026)**	**–0.43* (.040)**	1.00	
8. ASD characteristics[b]	**–0.47* (.023)**	0.11 (.632)	–0.10 (.635)	**–0.47* (.023)**	**–0.42* (.046)**	**–0.45* (.032)**	**0.66*** (< .001)**	1.00
9. Communication[c]	**0.62** (.002)**	–0.26 (.236)	–0.25 (.257)	**0.52* (.011)**	**0.57** (.005)**	**0.54** (.007)**	–0.07 (.748)	–0.27 (.211)
10. Daily living skills[d]	**0.61** (.002)**	–0.14 (.536)	–0.17 (.438)	**0.55** (.007)**	**0.59** (.003)**	**0.47* (.025)**	–0.21 (.342)	–0.23 (.290)
11. Socialization[e]	**0.60** (.003)**	–0.343 (.109)	–0.19 (.378)	**0.48* (.021)**	**0.49* (.019)**	**0.48* (.022)**	–0.28 (.203)	**–0.43* (.038)**
12. Vineland-3 ABC[f]	**0.68** (.001)**	–0.28 (.200)	–0.25 (.259)	**0.59** (.003)**	**0.61** (.002)**	**0.53** (.009)**	–0.227 (.297)	–0.40~ (.058)

Note. The bold values indicates significant values. Proportion scores used for responsivity and behavior management variables. C&I = complete and intelligible; NDW = number of different words; MLUm = mean length of utterance in morphemes; ASD = autism spectrum disorder; ABC-2 = Aberrant Behavior Checklist–Second Edition.

[a]ABC-2 total raw score. [b]SRS-2 total *T* score. [c]Vineland-3 Communication domain standard score. [d]Vineland-3 Daily Living Skills domain standard score. [e]Vineland-3 Socialization domain standard score. [f]Vineland-3 Adaptive Behavior Composite score. ~$p < .10$.

*$p < .05$. **$p < .01$. ***$p < .001$.

children with below-average levels of adaptive behavior. Additionally, across all dyads, when the other predictors were held constant, older children had higher levels of syntactic complexity than younger children. As with the other child language measures, there were no significant main effects of challenging behavior or parent sex on child syntactic complexity.

Behavior Management as a Predictor of Child Language

There was not a significant main effect of behavior management on child talkativeness (p = .672), lexical diversity (p = .163), or syntactic complexity (p = .971). These models also included challenging behavior, adaptive behavior, child age, and parent sex as predictors of the child language measures.

Aim 2 Models: Influence of Child Characteristics on Parental Behavior

The ICC for parental responsivity indicated that 68.2% of the variation was due to between-couples factors, whereas 31.8% was due to within-couple factors. For behavior management, 14.4% of the variation was due to between-couples factors, whereas 85.6% was due to within-couple factors. Table 9 presents the results of the MLM analyses for Aim 2. The behavior management variable was log-transformed to reduce positive skewness. Additionally, the Vineland-3 Socialization domain score was used in place of the Vineland-3 Adaptive Behavior Composite score to predict behavior management due to the significant negative correlation found between parental behavior management and the Vineland-3 Socialization domain score.

Prediction of Parental Responsivity

As expected, there was a significant main effect of child adaptive behavior on parental responsivity ($p <$ .001). Across all dyads, when the other predictors were held constant, children with above-average levels of adaptive behavior had parents who used higher rates of responsive behaviors compared to children with below-average levels of adaptive behavior. There was also a significant main effect of child age on parental responsivity ($p <$ .001). Across all dyads, when the other predictors were held constant, parents of older children demonstrated higher rates of parental responsivity than parents of younger children. There were no significant main effects of child challenging behavior or parent sex on parental responsivity.

Table 7. Spearman correlations for combined parental ratings and parent–child interaction variables (n = 46).

Variable	1	2	3	4	5	6	7	8
1. Responsivity	1.00							
2. Behavior management	–0.17 (.250)	1.00						
3. Child age	**0.34*** **(.019)**	0.17 (.271)	1.00					
4. Child total C&I utterances	**0.82***** **(< .001)**	0.14 (.356)	**0.40**** **(.006)**	1.00				
5. Child NDW	**0.83***** **(< .001)**	0.03 (.821)	**0.43**** **(.002)**	**0.93***** **(< .001)**	1.00			
6. Child MLUm	**0.73***** **(< .001)**	–0.06 (.703)	**0.48***** **(< .001)**	**0.79***** **(< .001)**	**0.91***** **(< .001)**	1.00		
7. Challenging behavior[a]	–0.19 (.218)	0.04 (.797)	–0.08 (.610)	–0.14 (.368)	–0.15 (.314)	–0.13 (.404)	1.00	
8. ASD characteristics[b]	**–0.39**** **(.007)**	0.16 (.277)	0.12 (.419)	–0.28~ (.063)	**–0.30*** **(.046)**	**–0.30*** **(.044)**	**0.58***** **(< .001)**	1.00
9. Communication[c]	**0.50***** **(< .001)**	–0.21 (.170)	–0.25 .100	**0.44**** **(.002)**	**0.51***** **(< .001)**	**0.49***** **(< .001)**	–0.10 (.499)	**–0.45**** **(.002)**
10. Daily living skills[d]	**0.51***** **(< .001)**	–0.18 (.239)	–0.17 (.258)	**0.49***** **(< .001)**	**0.57***** **(< .001)**	**0.43**** **(.003)**	–0.229 (.126)	**–0.44**** **(.002)**
11. Socialization[e]	**0.53***** **(< .001)**	**–0.35*** **(.016)**	–0.19 (.199)	**0.41**** **(.004)**	**0.46**** **(.001)**	**0.42**** **(.004)**	**–0.33*** **(.027)**	**–0.64***** **(< .001)**
12. Vineland-3 ABC[f]	**0.56***** **(< .001)**	–0.21 (.167)	–0.25 (.100)	**0.52***** **(< .001)**	**0.56***** **(< .001)**	**0.47**** **(< .001)**	–0.24 (.115)	**–0.59***** **(< .001)**

Note. The bold values indicates significant values. Proportion scores used for responsivity and behavior management variables. C&I = complete and intelligible; NDW = number of different words; MLUm = mean length of utterance in morphemes; ASD = autism spectrum disorder; ABC-2 = Aberrant Behavior Checklist–Second Edition.

[a]ABC-2 total raw score. [b]SRS-2 total T score. [c]Vineland-3 Communication somain standard score. [d]Vineland-3 Daily Living Skills domain standard score. [e]Vineland-3 Socialization domain standard score. [f]Vineland-3 Adaptive Behavior Composite score. ~p < .10.

*p < .05. **p < .01. ***p < .001.

Prediction of Behavior Management

There was a significant main effect of child adaptive behavior (i.e., socialization) on behavior management (p < .050). Across all dyads, when the other predictors were held constant, children with above-average levels of adaptive behavior in the Socialization domain had parents who used behavior management strategies proportionally less often compared to children with below-average levels of adaptive behavior in the Socialization domain. There was also a significant main effect of parent sex on behavior management (p < .050). In reference to the overall mean, when the other predictors were held constant, fathers demonstrated higher rates of behavior management strategies compared to mothers. There were, however, no significant main effects of challenging behavior or child age on parent behavior management.

Discussion

The current study was designed to examine relationships between parental responsivity and child language in mother–child and father–child dyadic interactions, as well as the ways in which child characteristics relate to both child language performance and parental behavior. The findings suggest that parental responsivity supports child language performance in terms of talkativeness and lexical diversity, with no discernable differences between child language performance in the mother–child and father–child interactions. There were also significant correspondences within families between mothers' and fathers' overall language use and responsiveness with the child, which is consistent with past research in families of neurotypical children (Tamis-LeMonda et al., 2012). This is the first time this association has been reported in families of children with FXS. Moreover, despite the significant correspondence between mothers' and fathers' language use, mothers were more talkative (i.e., had a higher total number of C&I utterances) and demonstrated greater lexical diversity (i.e., had a higher total NDW) compared to fathers. This finding is also consistent with past research that shows that mothers tend to provide more language input to their neurotypical children compared to fathers (Davidson & Snow, 1996; Pancsofar & Vernon-Feagans, 2006; Shapiro et al., 2021), but this is the first time that differences between maternal and paternal input during dyadic parent–child interactions have been reported in families of children with FXS. At the same time, however, these differences in maternal and paternal input did not lead to any significant differences in child language performance. However, because only

Table 8. Multilevel model results for Aim 1.

Variable	β (*SE*) Child talkativeness (total C&I utterances)	Child lexical diversity (NDW)[1]	Child syntactic complexity (MLUm)[1]
Fixed effects			
Intercept	**135.15***** **(9.79)**	**7.49***** **(0.40)**	**1.30***** **(0.03)**
Parental responsivity	**276.81***** **(57.89)**	**8.21***** **(1.98)**	0.31 (0.22)
Challenging behavior	0.19 (0.24)	−0.0004 (0.01)	−0.0001 (0.001)
Adaptive behavior	**2.51*** **(0.98)**	**0.16***** **(0.04)**	**0.01***** **(0.003)**
Child age	**20.60**** **(7.67)**	**1.07**** **(0.31)**	**0.09***** **(0.02)**
Parent sex	0.47 (3.82)	−0.06 (0.13)	0.01 (0.02)
Random effects			
Residual (σ^2_e)	632.12	0.68	0.01
Intercept (σ^2_{u0})	1890.14	3.42	0.01
Goodness-of-fit			
AIC	454.83	190.41	2.19
BIC	469.46	205.04	16.83

Note. The bold values indicates significant values. C&I = complete and intelligible; NDW = number of different words; MLUm = mean length of utterance in morphemes; AIC = Akaike information criterion; BIC = Bayesian information criterion.

[1]Variables were square-root-transformed to reduce positive skewness.

*p < .05. **p < .01. ***p < .001.

concurrent associations were examined in the current study, future investigations should continue to examine the potential differences between parents' overall language use, as well as any differences in responsivity and behavior management, to determine whether and how they influence the child's language development. Differential effects of parent input on child outcomes may emerge over time in longitudinal studies.

Some interesting differences emerged in the correlations between parental responsivity and child characteristics for mothers compared to fathers. For example, child ASD characteristics (which were independently rated by both mothers and fathers) were negatively related to parental responsivity for fathers but not for mothers, with fathers using a greater proportion of responsive behaviors with children who had fewer characteristics of ASD. Furthermore, child adaptive behavior in the Communication domain was positively related to parental responsivity for fathers but not for mothers, with fathers using a greater proportion of responsive behaviors with children who had higher levels of communication. Interestingly, child ASD characteristics were also negatively associated with all child language measures in the father–child interactions but not the mother–child interactions. These findings suggest that fathers may have more difficulty compared to mothers engaging in responsive behaviors with children who have greater levels of social impairment and lower levels of communication skills.

In contrast, child age was positively related to parental responsivity for mothers but not for fathers, with mothers using a greater proportion of responsive behaviors with older children. Past research in FXS has demonstrated a positive association between maternal responsivity and child rate of communication (Sterling et al., 2013), and child age in the current study was positively associated with child talkativeness, lexical diversity, and syntactic complexity in mother–child interactions. However, in the father–child interactions, child age was associated only with child syntactic complexity and not with talkativeness or lexical diversity. These findings suggest that mothers may be modifying their input to the child to a greater extent than fathers based on the child's age and developmental level. Future studies should investigate the differential contributions of child characteristics to parent behavior and child language in mother–child compared to father–child interactions to develop a better understanding of these potentially transactional relationships. Future studies should also consider how the relationships between these variables change over time.

In the multilevel models for Aim 1, parental responsivity was found to associate with child talkativeness and

Table 9. Multilevel model results for Aim 2.

Variable	β (*SE*)	
	Parental responsivity	Behavior management[a]
Fixed effects		
Intercept	**0.34****** (0.02)**	**–3.04****** (0.10)**
Challenging behavior	–0.0001 (0.0006)	0.001 (0.004)
Adaptive behavior[b]	**0.008****** (0.002)**	**–0.02* (0.01)**
Child age	**0.05****** (0.01)**	0.02 (0.08)
Parent sex	0.01 (0.01)	**0.25** (0.09)**
Random effects		
Residual (σ_e^2)	0.007	0.35
Intercept (σ_{u0}^2)	0.004	0.08
Goodness-of-fit		
AIC	–27.92	125.52
BIC	–15.12	138.32

Note. The bold values indicates significant values. AIC = Akaike information criterion; BIC = Bayesian information criterion.

[a]Variable was log-transformed to reduce positive skewness. [b]The Vineland-3 Socialization domain standard score was used to predict behavior management instead of the Vineland-3 Adaptive Behavior Composite score.

**p* < .05. ***p* < .01. ****p* < .001.

lexical diversity, but not syntactic complexity. Past research on parental responsivity in FXS has repeatedly failed to find an association between parental input and child syntax, and this has been true both for studies of naturalistic interactions (e.g., Brady et al., 2020; Komesidou et al., 2017) as well as studies of PILIs (e.g., McDuffie et al., 2018). For example, Komesidou et al. (2017), who examined the longitudinal trajectory of expressive syntax over 3 years in children with FXS, found significant syntactic growth over time, but maternal responsivity did not predict syntactic outcomes. Those authors suggested that perhaps more specific parental behaviors might contribute to growth of syntax and that their measure of maternal responsivity was potentially not specific enough. Additionally, certain responsive behaviors, such as requests for verbal compliance (e.g., questions such as, "What color is the truck?" or intonation prompts such as, "They are driving to the _____"), may only result in one- or two-word responses from the child (McDuffie et al., 2018). Parent use of other responsive behaviors, such as commenting on the child's focus of attention or recasting child communication acts, may not lead to observable or significant changes in the child's syntactic complexity, especially for young children. Future research is needed to investigate whether other parental behaviors may promote syntactic skills in children with FXS.

In the current study, parent behavior management did not predict any of the child language measures, demonstrating the importance of the specificity of parental input (e.g., comments and requests for verbal compliance compared to requests for behavioral compliance) in shaping child language development (e.g., McDuffie & Yoder, 2010). Past studies in families of children with FXS have repeatedly demonstrated that maternal responsivity predicts child language performance (Brady et al., 2014, 2020; Warren et al., 2010, 2017). Importantly, the findings of the current study not only replicate these previous findings in a new cohort of families but also demonstrate that *paternal* responsivity is also important for child talkativeness and lexical diversity, especially given that no significant differences were found in the child language measures between the mother–child and father–child interactions. These findings provide a more complete evaluation of the linguistic input of children who live in a two-parent household. Future studies should investigate how maternal and paternal behaviors change over time as the child develops and whether significant differences emerge between mothers and fathers that could differentially affect the child's communication. In particular, the role of paternal responsivity on child language performance during the school-age and adolescent years has not yet been explored in families of children with FXS.

In the multilevel models for Aim 2, there was not a significant main effect of child challenging behavior on either parental responsivity or behavior management. However, the children in the current study were generally very compliant during the dyadic play-based interactions and parents reported that challenging behaviors were more likely to occur during other interactions, particularly when demands were being placed on the child or there were unexpected changes in the child's routine. Parents' use of responsive behaviors may decrease during interactions when the child is demonstrating higher levels of challenging behaviors. Additionally, a more proximal measure of child challenging behavior (e.g., ratings of the child's behavior during an interaction) compared to a more distal measure (e.g., ABC-2 Total scores) may be more likely to relate to parent behavior. Therefore, future studies interested in investigating these associations should include additional interaction contexts as well as additional measures of the child's behavior.

Child adaptive behavior was also a significant predictor of parental responsivity such that children with higher levels of adaptive behavior had parents who were more responsive. This finding is similar to past research (Sterling et al., 2013). Moreover, child age was a significant predictor of parental responsivity, but not behavior management, with parents of older children demonstrating higher rates of responsivity. Sterling and Warren (2014) also found a positive association between child age and maternal responsivity in families of children with DS. Interestingly, adaptive behavior in the Socialization domain was a significant predictor of parental behavior management, with parents of children with lower levels of adaptive behavior in this domain implementing higher rates of behavior management. Parents of children with lower levels of social functioning may be more likely to use certain directives during interactions with their child to teach and encourage appropriate play. Moreover, even though there were no significant differences in the use of responsive strategies based on parent sex, there were differences in the use of behavior management strategies, such that fathers used a greater proportion of behavior management strategies compared to mothers. Future studies should examine how parental behavior management changes over time, whether fathers continue to use higher rates of behavior management compared to mothers, and how parental behavior management influences child developmental outcomes.

Given the association between child age and parental responsivity, the present findings suggest that parents of young children with FXS could benefit from interventions focused on increasing levels of responsive behaviors and that these interventions should start as early as possible to encourage child language development. Undoubtedly, parents of children who are more communicative will have an easier time implementing responsive behaviors, but responsive behaviors also serve to increase child engagement and participation in an interaction, thereby leading to improvements in the child's development. In the past decade, McDuffie, Abbeduto, and colleagues have published multiple studies examining the effects of PILIs on parent and child outcomes in families of children with FXS (Bullard et al., 2017; McDuffie et al., 2018; McDuffie, Machalicek, et al., 2016; McDuffie, Oakes, et al., 2016; Nelson et al., 2018; Oakes et al., 2015; Potter, Bullard, et al., 2022; Thurman et al., 2020). These interventions were designed to teach parents to use strategies that support their child's language development. In one study that included young boys with FXS (between the ages of 2 and 6 years) and their mothers, mothers increased their use of responsive strategies, including comments and prompts for child communication (e.g., requests for verbal compliance) and the children showed increases in their prompted communication acts (McDuffie, Oakes, et al., 2016).

Other studies of a PILI for school-age children and adolescents with FXS have also shown improvements in parent use of responsive strategies and child language performance (McDuffie et al., 2018; McDuffie, Machalicek, et al., 2016; Nelson et al., 2018; Thurman et al., 2020). In these studies, the parent–child interaction context was shared storytelling using wordless picture books and parents were taught to (a) model developmentally appropriate story-related vocabulary and grammar, (b) expand (i.e., recode) child communication acts, (c) ask *wh*-questions, and (d) use intonation prompts (i.e., fill-in-the-blank statements). Parents were able to successfully learn and implement these strategies independently over the course of the intervention, and there were associated improvements in child participation and language. Collectively, these studies demonstrate that parental responsiveness is important for child outcomes in FXS from early childhood through late adolescence and that parents are able to successfully implement targeted strategies to children who vary widely in both age and developmental level.

Limitations and Future Directions

There are some notable limitations to this study. First, only males with FXS were included and the sample size is relatively small and homogeneous in terms of race/ethnicity and socioeconomic status. Additionally, the measures of child language are from the same interactions being used to ascertain levels of parental responsivity. Future studies should incorporate both males and females with FXS as well as additional external or distal measures of child language, especially other measures of syntax. Moreover, including additional parent–child interaction contexts, such as shared book reading and unstructured naturalistic activities (e.g., getting ready for school, eating dinner), would potentially provide more representative information about the nature of the parent–child relationship and the ways in which parental behavior influences child behavior and communication throughout the day in various settings. Another limitation is that the current findings describe concurrent associations and not longitudinal ones. Future studies should examine changes in these bidirectional parent–child associations over time to see how parents modify their behavior to adapt to the child's development.

Conclusions

The findings from the current study demonstrate that both maternal and paternal responsivity are positively associated with child language performance for young boys with FXS. Interestingly, there were no significant differences within families between mothers' and fathers' use of responsive behaviors. Future studies should investigate whether there are differences in maternal and paternal behavior in dyadic compared to triadic (i.e., mother–father–child) interactions in these families. This study also provides preliminary evidence that certain child attributes (e.g., ASD characteristics) may differentially affect maternal versus paternal responsivity, which warrants further investigation. Finally, the associations between both child age and adaptive functioning with parental responsivity support the use of PILIs in families of children with FXS to increase parents' use of responsive strategies that target improvements in child communication.

Author Contributions

Sarah Nelson Potter: Conceptualization (Lead), Data curation (Lead), Formal analysis (Lead), Funding acquisition (Supporting), Investigation (Lead), Methodology (Lead), Project administration (Lead), Writing – original draft (Lead), Writing – review & editing (Lead). **Danielle Harvey:** Data curation (Supporting), Formal analysis (Supporting), Writing – review & editing (Supporting). **Audra Sterling:** Conceptualization (Supporting), Methodology (Equal), Project administration (Supporting), Writing – review & editing (Supporting). **Leonard Abbeduto:** Conceptualization (Supporting), Funding acquisition (Lead), Methodology (Equal), Project administration (Lead), Writing – review & editing (Supporting).

Data Availability Statement

The data sets used and/or analyzed during this study are available from the corresponding author on reasonable request.

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