Title
An Examination of Family Dynamics, Parental Responsivity, and Child Communication in Fragile X Syndrome

Author
Potter, Sarah Nelson

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An Examination of Family Dynamics, Parental Responsivity,
and Child Communication in Fragile X Syndrome

By

SARAH NELSON POTTER
DISSERTATION

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Approved:

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Leonard Abbeduto, Chair

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Danielle Harvey

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David Hessl

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Aubyn Stahmer

Committee in Charge

2021

Dedication

To my sister, Erica, who taught me the importance of having a voice,

and the joys of caring for those you love.

Abstract

Children with fragile X syndrome (FXS) have significant delays in many domains of functioning. Biological mothers of children with FXS are at an increased genetic risk for experiencing cognitive, physical, and mental health challenges. Parental mental health challenges and stress are often associated with reduced marital cohesion and satisfaction, which is likely to spill over and negatively affect the parent-child relationship for both mothers and fathers. Past research shows that parentally responsive behavior positively influences language development in both neurotypical children and children with intellectual and developmental disabilities, including those with FXS. However, the majority of past studies on parent-child interactions, and families of children with FXS more generally, have focused exclusively on the mother-child relationship. Therefore, very little is known about fathers in these families, including their well-being and their role in the child's development. The current dissertation fills this gap by examining multiple features of the family environment, including the mother-father relationship, the mother-child relationship, and the father-child relationship in 23 families of children with FXS. Study 1 examined parental and couple well-being and associations between child functioning and these domains. Mothers and fathers independently completed questionnaires regarding their mental health, parenting stress, couples satisfaction, and dyadic coping in their relationship. Parents also independently completed questionnaires regarding their child's challenging behaviors and symptoms of autism spectrum disorder (ASD). One parent also completed an interview about the child's adaptive functioning. Results from this study indicated that mothers and fathers in these families experienced elevated levels of clinically significant mental health challenges and parenting stress compared to levels reported in the general population. However, despite these challenges, the majority of both mothers and fathers reported average to above average levels of couple well-being (i.e., couples satisfaction and dyadic coping). Multilevel models indicated that higher levels of parenting stress predicted lower levels of both couples satisfaction and dyadic coping. Additionally, multilevel models indicated that higher levels of child challenging behavior predicted higher levels of mental health challenges and parenting stress as well as lower levels of couples satisfaction and dyadic coping. Moreover, the parents of children with higher levels of adaptive functioning reported lower levels of parenting stress and higher levels of couples satisfaction. There were no significant differences in these relationships between mothers and fathers. Study 2 examined relationships among maternal responsivity, paternal responsivity, and parental and couple functioning. Mothers and fathers separately engaged in 12-minute dyadic play-based interactions with their child. These interactions were recorded via secure teleconferencing in the families' homes. Recordings were transcribed and coded for the presence of parentally responsive behaviors and behavior management strategies. Results indicated significant correspondences between mothers' and fathers' frequencies and rates of responsive behaviors during the parent-child dyadic interactions. However, overall, mothers used comments more frequently than fathers, and fathers used a higher proportion of behavior management strategies compared to mothers. Multilevel models indicated that higher levels of parenting stress predicted lower rates of parental responsivity and higher rates of behavior management, but these effects were only marginally significant. Couples satisfaction did not predict either category of parent behavior. Study 3 examined relationships among maternal responsivity, paternal responsivity, and child language; it also examined relationships between child characteristics (i.e., challenging behavior, ASD symptoms, and adaptive behavior) and child language as well as parent behavior. Measures of parent and child language were obtained from the transcripts of the mother-child and father-child interactions. The measures were talkativeness, lexical diversity, and syntactic complexity. Multilevel models indicated that both maternal and paternal responsivity were positively associated with child talkativeness and lexical diversity. Parental responsivity, however, was not associated with child syntactic complexity. Findings also indicated that older children and children with higher levels of adaptive behavior had parents who used higher rates of responsive behaviors; these children also had higher levels of talkativeness, lexical diversity, and syntactic complexity. Additionally, fathers used higher rates of behavior management strategies compared to mothers, and this type of parental behavior was not associated with child language performance. The findings of this dissertation advance our understanding of family relationships in families of children with FXS as well as features of dyadic relationships within families that promote optimal outcomes.

An Examination of Family Dynamics, Parental Responsivity,
and Child Communication in Fragile X Syndrome

The three papers that comprise this dissertation sought to examine multiple features of the family environment in families of young boys with fragile X syndrome (FXS). FXS is the leading inherited cause of intellectual disability (Crawford et al., 2001) and results from an expansion in the *FMR1* gene located on the X chromosome. Individuals with FXS experience a wide range of developmental impairments, including delays in language (Abbeduto et al., 2007), symptoms of autism spectrum disorder (ASD; Abbeduto et al., 2019) and attention deficit hyperactivity disorder (ADHD; Chromik et al., 2015), heightened levels of anxiety (Cordeiro et al., 2011), and increased rates of challenging behaviors (Hatton et al., 2002). Biological mothers of children with FXS, who are carriers of either the *FMR1* premutation or full mutation, are genetically predisposed to experiencing elevated levels of mental health challenges, including depression and anxiety (Bailey et al., 2008). Moreover, given the challenges associated with raising a child with significant impairments, these mothers are also likely to experience elevated levels of parenting stress (Abbeduto et al., 2004). Past research also suggests that mothers of children with FXS may experience reduced marital satisfaction (Baker et al., 2012). Additionally, child characteristics, including challenging behaviors and symptoms of autism spectrum disorder (ASD), have been shown to negatively affect parental and couple well-being (e.g., Abbeduto et al., 2004; Baker et al., 2012; Fielding-Gebhardt et al., 2020; Johnston et al., 2003 Lewis et al., 2006; McCarthy et al., 2006). Many past studies of parent-child relationships in FXS have demonstrated the importance of responsive parenting for a range of developmental outcomes in children with FXS (e.g., Brady et al., 2014; Warren et al., 2017). However, the majority of past studies have focused on the mother-child relationship or the effects of the child's characteristics on aspects of maternal well-being. Therefore, very little is known about fathers in these families.

The papers that follow describe three studies that build upon past research in families of children with FXS by examining features of the entire family system, including the mother-father relationship, the mother-child relationship, and the father-child relationship. Study 1 examined relationships between maternal and paternal well-being and their associations with couple functioning, as well as the ways in which characteristics of the child relate to these domains of parent functioning. Study 2 examined how these parental and couple factors related to parental responsivity in both mother-child and father-child dyadic interactions with the child. Finally, Study 3 examined the differences between maternal and paternal behavior during parent-child dyadic interactions and their relationships to child language performance. Study participants were 23 families of young boys with FXS. All data were collected in the family home at a distance through video teleconferencing and online questionnaires, which had both cost and logistical benefits, and also allowed for uninterrupted data collection during the COVID-19 pandemic.

Study 1 was designed to examine relationships among maternal and paternal mental health challenges and parenting stress as well as couple functioning in families of young boys with FXS. In addition, Study 1 examined relationships between characteristics of the child and parent and couple well-being. The results of Study 1 suggest that mothers *and* fathers in these families experience clinically significant levels of mental health challenges and elevated rates of parenting stress relative to the general population. However, the majority of parents reported average to above average levels of couples satisfaction and dyadic coping, indicating that the mother-father relationship may be a source of strength or resilience for these parents. Parents of children with higher levels of challenging behaviors experienced greater levels of mental health challenges and parenting stress, as well as lower levels of both couples satisfaction and dyadic coping. Moreover, parents of children with higher levels of adaptive behavior reported less parenting stress and greater couples satisfaction.

Study 2 was designed to examine relationships between parent and couple characteristics (i.e., those examined in Study 1) and parent behavior (i.e., responsivity and behavior management) during mother-child and father-child dyadic interactions in the same families of young boys with FXS. Past research has demonstrated that parent mental health challenges and stress are associated with lower levels of responsiveness in interactions with the child (e.g., Sterling et al., 2013; Wheeler et al., 2007). Additionally, mothers and fathers who have stable and healthy relationships are more likely to have positive and responsive parent-child relationships and thereby, children with more optimal outcome (e.g., Greenlee et al., 2021). The first aim of Study 2 was to examine relationships among maternal responsivity, paternal responsivity, and parent individual well-being (i.e., mental health challenges and parenting stress). The second aim of Study 2 was to examine relationships among maternal responsivity, paternal responsivity, and couple well-being (i.e., couples satisfaction and dyadic coping). The results of this study indicated that mothers and fathers use similar rates of responsive behaviors with their child, but that fathers use higher rates of behavior management strategies compared to mothers (i.e., they are more directive during play interactions). Additionally, parenting stress predicted lower rates of parental responsivity and higher rates of behavior management, but these effects were only marginally significant. Couples satisfaction, however, which was highly negatively skewed in this sample of parents, did not predict parent behavior during dyadic parent-child interactions.

Study 3 was designed to examine relationships between parent behavior and child language performance in mother-child and father-child dyadic interactions, as well as relationships between child characteristics and both parent behavior and child language performance. Past research demonstrates that parental responsivity positively influences language development in neurotypical children (e.g., Landry et al., 2006) and children with intellectual and developmental disabilities (e.g., Brady et al., 2009; Warren & Brady, 2007), including FXS (e.g., McDuffie et al., 2018; Warren et al., 2010), ASD (e.g., Haebig et al., 2013; McDuffie & Yoder, 2010), and Down syndrome (e.g., Yoder & Warren, 2004). However, nearly all of these studies have focused exclusively on the mother-child relationship, or maternal responsivity. Therefore, very little is known about how paternal behavior compares to maternal behavior or how paternal behavior relates to child outcomes. Results of Study 3 indicated that both maternal and paternal responsivity were positively associated with child language performance, including talkativeness and lexical diversity. Parental responsivity was not associated with child syntactic complexity, and parental behavior management was not associated with any of the child language measures. Additionally, mothers and fathers of older children and children with higher levels of adaptive behavior were found to use higher rates of responsive behavior.

Across the three studies, we found many similarities between mothers and fathers, both in their levels of individual and couple well-being, as well as in their behavior during dyadic interactions with their child. These studies also provide evidence that families of children with FXS would likely benefit from interventions focused on reducing parent stress, such as Mindfulness Based Stress Reduction (MBSR; Neece, 2014). Other interventions that are likely to improve family functioning include parent-implemented interventions focused on decreasing child challenging behaviors (e.g., Hall et al., 2020) as well as those focused on increasing levels of parental responsivity (e.g., McDuffie et al., 2018). Child and family-based services that include both mothers and fathers would likely lead to better outcomes for all family members (Fox et al., 2015; Wang et al., 2006). The data from this dissertation provide a rationale for future studies investigating family relationships in families of children with FXS and other intellectual and developmental disabilities, particularly those focused on understanding child developmental trajectories in the context of the family environment as well as bidirectional relationships between parent and child functioning.

Participants

Fathers, biological mothers, and male children with FXS between 3;0 and 7;11 years of age were recruited to participate in the broader study on which the three papers that comprise this dissertation are based. Families were recruited through (1) a database of previous research participants, (2) the MIND Institute IDDRC Clinical Translational Core's research participant registry, (3) a listserv and study information page facilitated by the National Fragile X Foundation (NFXF), and (4) the NFXF's FORWARD Registry and Database. Eligibility criteria were a) the child lived at home with both parents, b) English was the primary language spoken in the home, and c) the child had no uncorrected sensory or motor impairments that would limit his ability to participate in the study. 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, Davis in advance of recruitment, and both parents provided informed consent electronically via REDCap (Research Electronic Data Capture; Harris et al., 2019; Harris et al., 2009).

A total of 37 families of male children with FXS between the ages of 3;0 and 7;11 years were screened for participation in the current study between October 2019 and April 2021. One family did not meet eligibility criteria, four families declined participation, and seven families were lost to follow-up. Twenty-five families completed the informed consent process, but two families left the study prior to data collection due to the COVID-19 pandemic. Therefore, the current study includes a total of 69 participants: 23 fathers (22 biological fathers and one stepfather), 23 biological mothers, and 23 male children with FXS. Participant characteristics are presented in Table 1. A majority of the participants identified as white and not Hispanic or Latinx. Twenty mothers were carriers of the *FMR1* premutation, two were carriers of the *FMR1* full mutation, and one had not been tested. A majority of both mothers and fathers in the study had at least a bachelor's degree and parent-reported household income indicated that most families were relatively well-resourced. All families resided in North America, with 13 U.S. states and two Canadian provinces represented. Data were collected between December 2019 and July 2021; therefore, the majority of families were tested during the COVID-19 pandemic. Only two families completed their participation in the study prior to the first community-diagnosed case in California on February 23, 2020.

Procedures

Participation in the study involved multiple calls with an examiner via video teleconferencing, the completion of online questionnaires, and an interview. Mothers and fathers separately engaged in play-based dyadic interactions with their child on separate days of the study. Mothers and fathers also independently completed multiple questionnaires about their individual well-being, couple functioning, and child behavior. One parent completed an interview about the child.

Skype for Business and Zoom were the preferred teleconferencing platforms for securely connecting with the participating families. These platforms were approved by UC Davis Health IT and Research Compliance. With both Skype for Business and Zoom, calls were hosted through the examiner's UC Davis account and participants joined the calls as a guest. Using these procedures, the examiner was able to securely record each call and notified the participants when the recording was being started.

All questionnaire data were collected and managed using REDCap electronic data capture tools hosted at the University of California, Davis (Harris et al., 2019; Harris et al., 2009). REDCap is a secure, web-based software platform designed to support data capture for research studies, providing 1) an intuitive interface for validated data capture; 2) audit trails for tracking data manipulation and export procedures; 3) automated export procedures for seamless data downloads to common statistical packages; and 4) procedures for data integration and interoperability with external sources.

Additionally, a unique study SharePoint page was created for each family so that they could easily connect to the study teleconferencing calls and access their REDCap questionnaires. Prior to data collection, each family participated in a training call with the lead researcher. The purpose of this initial call was to orient the family to the study procedures and technology that would be used for the duration of the project. See Figure 1 for additional details regarding study design and procedures.

Table 1

Family Demographic Characteristics

Individual Characteristics	Child	Mother	Father
Age (years)			
Mean (*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/Latinx	20 (87%)	20 (87%)	19 (83%)
Hispanic/Latinx	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%)
Previous or current psychiatric diagnosis[1]	7 (30%)		3 (13%)
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%)	
Additional siblings in family (*n*, %)[2]			
0		3 (13%)	
1		13 (56%) [4]	
2		5 (22%) [4]	
3		2 (9%)	

Note. The individual percentage values are rounded and may not total 100%. [1]According to parent report. [2]Number of additional siblings in family with a disability noted in brackets.

Measures

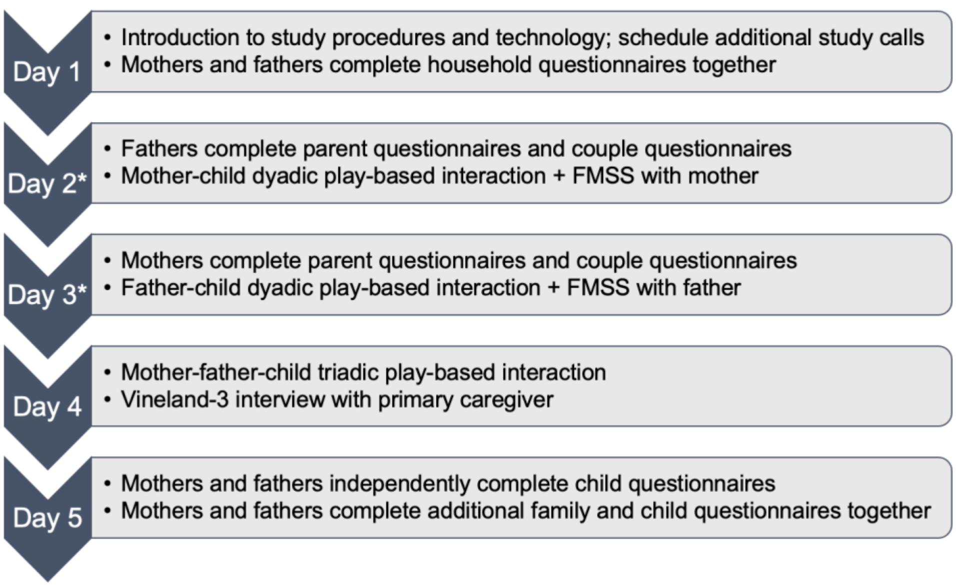
Individual-level Parent Measures

Parents independently completed two questionnaires via REDCap to assess their mental health challenges and views on parenting.

Symptom Checklist-90-Revised (SCL-90-R; Derogatis, 1994) – The SCL-90-R is a 90-item scale that measures mental health symptoms along the following dimensions: Somatization, Obsessive-Compulsive, Interpersonal Sensitivity, Depression, Anxiety, Hostility, Phobic Anxiety, Paranoid Ideation, Psychoticism, and additional symptoms, yielding a Positive Symptom Total, a Positive Symptom Distress Index, and a Global Severity Index. Lower scores indicate lower levels of mental health challenges. T-scores from each dimension and the Global Severity Index (representing overall mental health challenges) were used in analyses. The scale takes approximately 15 minutes to complete.

Figure 1

Study Design



Note. Days 2 and 3 were counter-balanced across families.

Parenting Stress Index—Fourth Edition, Short Form (PSI-4-SF; Abidin, 2012) – The PSI-4-SF is a 36-item scale that measures parenting stress in the domains of anxiety, mood, relationships, attachment, and family mental health and functioning. Like the 120-item PSI, the short form provides scores on the following subscales: (1) Parental Distress, (2) Parent-Child Dysfunctional Interaction, and (3) Difficult Child. Lower scores on these subscales indicate lower levels of parenting stress. T-scores and percentiles from these dimensions as well as the Total Stress score were used in analyses. This questionnaire takes approximately 10 minutes to complete.

Couple-level Parent Measures

Parents independently completed two questionnaires via REDCap to assess aspects of their couple functioning.

Couples Satisfaction Index (CSI-32; Funk & Rogge, 2007) – The CSI-32 is a 32-item scale that measures satisfaction in the couple's relationship, with higher total scores indicating higher satisfaction. Item-response theory was used to develop the CSI-32 from a set of 180 relationship satisfaction items administered to over 5,000 individuals. Compared to previous measures of relationship satisfaction, the CSI-32 demonstrates higher precision and has strong internal consistency and construct validity. Scores on the CSI-32 range from 0 to 161. Scores below 104.5 indicate notable relationship dissatisfaction. This scale takes approximately 10 minutes to complete.

Dyadic Coping Inventory (DCI; Bodenmann, 2008; Ledermann et al., 2010) – The DCI is a 37-item scale that measures perceived communication and coping that occurs in relationships when one or both partners are experiencing stress. In this measure, dyadic coping is assessed as a multidimensional construct that includes the following components: supportive, delegated, negative, and joint (common) coping. The DCI helps to assess an individual's perceptions about both the quality and quantity of the partner's support in the dyadic relationship. Scores on the DCI range from 35 to 175. Scores below 111 indicate below average dyadic coping, whereas scores above 145 indicate above average coping. This scale takes approximately 10 minutes to complete.

Child Measures

Parents independently completed two questionnaires via REDCap to assess child challenging behavior and symptoms of ASD. Additionally, one parent completed an interview to assess the child's level of adaptive functioning.

Aberrant Behavior Checklist, 2nd Edition (ABC-2; Aman & Singh, 2017) – The ABC-2 is a 58-item scale developed to assess challenging behaviors of individuals with developmental disabilities in several domains. For this study, subscale scoring based on the revised FXS-specific factor structure from Sansone et al. (2012) was used. The following factors are included in the FXS-specific subscale scoring: Irritability, Socially Unresponsive/Lethargic, Stereotypy, Hyperactivity, Inappropriate Speech, and Social Avoidance. Total raw scores for the FXS-specific factor structure scoring range from 0 to 165 and were used in the present analyses (Sansone et al. (2012) omitted three items from the original ABC due to weak loadings in the exploratory factor analysis). This checklist takes approximately 10 minutes to complete.

Social Responsiveness Scale, 2nd Edition (SRS-2; Constantino & Gruber, 2012) – The SRS-2 is a 65-item scale used to assess social impairments commonly associated with ASD. 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. On the SRS-2, the following subscales are included in addition to a total score: Social Awareness, Social Cognition, Social Communication, Social Motivation, and Restricted Interests and Repetitive Behavior. DSM-5 compatible subscale scores include a Social Communication and Interaction (SCI) score and a Restricted Interests and Repetitive Behavior (RRB) score. SCI, RRB, and Total T-scores were used in analyses. The scale takes approximately 15 minutes to complete.

Vineland Adaptive Behavior Scales, 3rd Edition (Vineland-3; Sparrow, Cicchetti, & Saulnier, 2016) – The Vineland-3 measures adaptive behavior across the following domains: Communication, Socialization, Daily Living Skills, Motor Skills, and Maladaptive Behavior. For this study, only the Communication, Socialization, and Daily Living Skills domains were administered. The Vineland-3 was administered as an interview by a trained examiner using Q-Global, a web-based platform for online administration. The child's primary caregiver (as reported by the parents) was interviewed over the phone or via a secure teleconferencing platform (i.e., Skype for Business or Zoom). The Vineland-3 is a norm-based instrument with a mean standard score of 100 and a standard deviation of 15. The Adaptive Behavior Composite score as well as the Communication, Daily Living Skills, and

Socialization domain standard scores were reported and used in analyses. The Vineland-3 interview takes approximately one to two hours to complete.

The SCL-90-R, PSI-4-SF, CSI-32, DCI, ABC-2, and SRS-2 are traditionally paper-and-pencil measures. They were modified so that they could be completed in packages (i.e., individual parent measures, couple measures, child measures) as online surveys via REDCap during different days of the study (see Figure 1).

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 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 minutes. Families were told that they could also include any 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 their camera and muted their 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 SALT (Systematic Analysis of Language Transcripts; 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. Following this, the primary transcriber finalizes the transcript based on the second transcriber's feedback. These procedures yield 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 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 from transcripts of 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 along with 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 three second pause or a child communication act was coded. See Table 2 for definitions and examples of the parental behavior codes. 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 parent utterances in the transcript. For example, a parent who had 144 responsive utterances, 17 behavior management utterances, and 227 total utterances would have a responsivity proportion of 144/227 = 0.63 and a behavior management proportion of 17/227 = 0.07.

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 resolved via consensus coding. Inter-observer agreement of total scores for parental responsivity and behavior management codes was based on a random sampling of approximately 20% of the sessions. Two-way random intra-class correlation coefficients (ICCs) were .994 for the parental responsivity composite and .937 for the behavior management composite. Additionally, ICCs for the subcategories of parental responsivity and behavior management 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 two occurrences per dyadic interaction.

The three papers that comprise this dissertation utilized data from the broader study described above. Brief summaries of these procedures and measures will be included in the papers that follow.

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	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!"

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Study 1: An Examination of Mental Health Challenges, Parenting Stress, and Features of the Couple Relationship in Parents of Children with Fragile X Syndrome

Abstract

Individuals with fragile X syndrome (FXS) have significant delays in cognition and language, as well as symptoms of autism spectrum disorder, anxiety, and challenging behaviors including hyperactivity and aggression. Biological mothers of children with FXS, who are themselves *FMR1* premutation or full mutation carriers, are at elevated risk for mental health challenges in addition to experiencing the stress associated with parenting a child with significant disabilities. However, little is known about fathers in these families, including the ways in which parental well-being influences the mother-father relationship and the impact of child characteristics on paternal and couple functioning. The current study examined features of, and relationships between, parental well-being, couple well-being, and child functioning in 23 families of young boys with FXS. Results suggest that both mothers and fathers in these families experience clinically significant levels of mental health challenges and elevated rates of parenting stress relative to the general population. Findings also indicate that the mother-father relationship may be a source of strength that potentially buffers against some of the daily stressors faced by these families. Additionally, parents who reported less parenting stress also had higher couples satisfaction and dyadic coping. Finally, parents of children with less severe challenging behaviors exhibited fewer mental health challenges, less parenting stress, and higher levels of both couples satisfaction and dyadic coping, and parents of children with higher levels of adaptive behavior also reported less parenting stress and higher couples satisfaction. Overall, this study provides evidence that families of children with FXS need access to services that not only target improvements in the child's functioning, but also ameliorate the parents' levels of stress. Family-based services that include both mothers and fathers would lead to better outcomes for all family members.

Keywords: fragile X syndrome, mental health, parenting stress, couple relationships, parent-child relationships

Children with fragile X syndrome (FXS), the leading inherited cause of intellectual disability (ID; Crawford et al., 2001), demonstrate delays in multiple domains of spoken language (Abbeduto et al., 2007). In addition, these children also present with increased rates of challenging behaviors, symptoms of autism spectrum disorder (ASD), inattention, hyperarousal, and anxiety (Hagerman et al., 2017). Mothers of children with FXS, who transmit the full mutation of the *FMR1* gene which causes FXS to their children, are themselves either carriers of the *FMR1* premutation or the full mutation. Both the *FMR1* premutation and the full mutation are associated with a multitude of physical, mental health, and cognitive challenges (Hagerman et al., 2018; Hartley, Seltzer, et al., 2011), which when compounded with the characteristics of the child with FXS, could negatively impact mother-child interactions and thus, child development. In addition, both mothers and fathers of children with FXS are likely to experience heightened levels of parenting stress due to characteristics of their children (McCarthy et al., 2006), which may impede their ability to engage with their child in the sustained and productive interactions needed to facilitate the child's development. Elevated parenting stress, which is likely given the challenging behaviors of individuals with FXS, is also likely to have a negative impact on the marital relationship for parents of children with FXS, which may further exacerbate the relationship between parents and their children (Peltz et al., 2018).

Unfortunately, the majority of past studies on parenting in FXS have focused exclusively on the mother-child dyad. In doing so, these studies have neglected to consider the role that fathers play in the development of the child or how features of the broader family environment may influence maternal or paternal behavior and child outcomes. The current study was designed to examine the broader family environment in families of young children with FXS, including maternal and paternal well-being, features of the mother-father relationship, and relationships between child characteristics and parent and couple well-being. A better understanding of parent and couple well-being in families of children with FXS, as well as the ways in which child characteristics influence these domains, will provide the foundation for developing interventions and services focused on improving outcomes for all family members.

The Impact of *FMR1* Mutation Phenotypes on the Family System

FXS is an X-linked disorder that results from an expansion of a cytosine-guanine-guanine (CGG) sequence in the promoter region of the *FMR1* gene, located at Xq27.3, from the typical 35 or so repeats to greater than 200 repeats (Oostra & Willemsen, 2003). Individuals with more than 200 CGG repeats have the full mutation, whereas individuals with 55 to 200 CGG repeats are considered premutation carriers. The full mutation typically leads to hypermethylation and transcriptional silencing of *FMR1*, causing a deficiency in, or absence of, the gene's associated protein, FMRP (fragile X mental retardation 1 protein), which is critical for early brain development, including synaptic protein synthesis and plasticity as well as experience-dependent learning (Bhakar et al., 2012; Hagerman et al., 2017; Hagerman et al., 2005). In contrast, the premutation typically involves elevated levels of *FMR1* mRNA, which leads to RNA toxicity. RNA toxicity is associated with reduced neuronal function, oxidative stress, chronic DNA damage repair changes, and ultimately the development of fragile X-associated tremor/ataxia syndrome (FXTAS; Hagerman et al., 2018; Wheeler et al., 2014) and other co-occurring physical and behavioral health challenges (described subsequently).

Because it is inherited, the presence of FXS in a family has far-reaching intergenerational effects, offering a unique opportunity to investigate the ways in which multiple family subsystems influence child outcomes. Nearly all males with FXS have ID (Hessl et al., 2009), and many also experience a variety of other conditions, including hyperactivity, attention problems, anxiety, symptoms of ASD, aggressive and self-injurious behaviors, and abnormal sensory processing (Bailey et al., 2008; Raspa et al., 2018). Language is also significantly impaired in individuals with FXS, with some domains affected to an even greater extent than would be expected based upon their level of cognitive functioning (Finestack & Abbeduto, 2010; Kover & Abbeduto, 2010; Kover et al., 2012). The combination of cognitive and psychiatric impairments in boys with FXS reduce the likelihood that they will be able to engage in successful and productive interactions critical for the development of cognitive, language, and social skills (Abbeduto et al., 2007).

The biological mothers of children with FXS are most often carriers of the *FMR1* premutation, although some also have the full mutation which causes FXS. Full mutation mothers are at an increased risk for experiencing mental health challenges, including anxiety and depression, as well as social deficits, including avoidance and withdrawal (Bailey et al., 2008; Franke et al., 1998; Freund et al., 1993; Hagerman et al., 2018; Hartley, Seltzer, et al., 2011; Keysor & Mazzocco, 2002; Wheeler et al., 2014). Women with the *FMR1* premutation may also experience deficits in executive functioning, memory, and language (Klusek et al., 2021; Sterling, Mailick, et al., 2013; Wheeler et al., 2014). Moreover, cognitive functioning is variable in women with FXS, ranging from severe impairment to above average, with most of these women demonstrating IQs in the range of average to slightly below average intelligence (Bartholomay et al., 2019). However, even some with average-range intelligence can have a learning disability and/or deficits in executive functioning and attention (Hall & Berry-Kravis, 2018). These cognitive phenotypic features of premutation and full mutation mothers are significant given that low maternal IQ is a risk factor for poorer child outcomes (Hooper et al., 1998; Sterling, Warren, et al., 2013).

Unfortunately, the mental health conditions that are experienced by both premutation and full mutation mothers of children with FXS can also be exacerbated by the stress they are likely to experience as a result of raising a child with significant challenges and impairments (Abbeduto et al., 2004; Hagerman & Hagerman, 2004). Furthermore, maternal depression and anxiety are associated with disrupted marital cohesion and decreased couples satisfaction (Baker et al., 2012; Essex et al., 2003). Of course, disruptions or problems in the marital relationship also affect paternal well-being, which may in turn negatively influence the quality of the relationship between father and child (England & Sim, 2009). Overall, poor parental and marital functioning have been repeatedly shown to contribute to negative child outcomes in neurotypical children (Hanington et al., 2012; Masarik & Conger, 2017; Peterson & Zill, 1986).

Many previous studies have found that child characteristics, partner characteristics, and features of the marital relationship differentially affect mothers and fathers of children with disabilities (e.g., Bristol et al., 1988; Dabrowska & Pisula, 2010; Hartley et al., 2016; Hastings, 2003). For example, in families including children with ASD, fathers are likely to be negatively affected by the child's challenging behaviors to an even greater extent than are mothers (Davis & Carter, 2008). The same may be true in families affected by FXS given the symptom overlap between FXS and ASD (Abbeduto et al., 2014). Moreover, maternal anxiety and depression—as well as the mother's parenting stress—are also likely to take a significant toll on fathers in these families (Hartley, Seltzer, Hong, et al., 2012), which could spill over and negatively affect the father-child relationship. McCarthy et al. (2006) found that both mothers and fathers of children with FXS reported high levels of stress, but that the predictors of stress differed between mothers and fathers with the strongest predictor of maternal stress being marital satisfaction and the strongest predictor of paternal stress being the child's level of adaptive skills.

Very little else is known about fathers of children with FXS given that the majority of past studies have focused on the mother-child dyad. However, including both mothers and fathers in behavioral therapies and health care services positively contributes to a child's success, especially for young children (Fox et al., 2015; Wang et al., 2006). In order to maximize treatment gains for children with FXS and to improve well-being for the entire family system, researchers and clinicians need to develop a greater understanding of the challenges faced by families affected by FXS. This understanding will inform services and interventions for these families.

Current study

The current study was designed to examine multiple features of the family environment, including maternal and paternal mental health, stress associated with parenting, aspects of couple functioning, and relationships between child characteristics and these parental domains. The first aim was to examine mental health challenges and parenting stress in biological mothers of children with FXS, who are carriers of the *FMR1* premutation or the full mutation. We hypothesized that these mothers, compared to the general population, would report elevated levels of mental health challenges and parenting stress (e.g., Hagerman et al., 2018; Hartley, Seltzer, et al., 2011; Wheeler et al., 2007). The second aim was to examine mental health challenges and parenting stress in fathers of children with FXS and compare paternal and maternal mental health challenges and parenting stress. We hypothesized that fathers whose partners reported experiencing elevated levels of mental health challenges and parenting stress would themselves report elevated levels of mental health challenges and parenting stress compared to the general population based on past findings in families of children with ASD, Down syndrome, and FXS (Hartley, Seltzer, Head, & Abbeduto, 2012). The third aim was to examine relationships between aspects of the couple relationship (i.e., couples satisfaction and dyadic coping) and mothers' and fathers' mental health challenges and parenting stress. We hypothesized that couples satisfaction and dyadic coping would be negatively related to mental health challenges and parenting stress for both mothers and fathers (Kersh et al., 2006). Finally, the fourth aim was to examine relationships between child characteristics (i.e., challenging behaviors, ASD symptoms, and adaptive behavior) and parental individual well-being (i.e., mental health challenges and parenting stress) and couple well-being (i.e., couples satisfaction and dyadic coping). We hypothesized that children with higher levels of behavior problems and ASD symptoms, as well as lower levels of adaptive behavior, would have parents who endorsed lower levels of individual well-being (Abbeduto et al., 2004) and couple well-being (Baker et al., 2012) with fathers being affected by child characteristics to a greater extent than mothers (McCarthy et al., 2006).

Method

Procedures and Measures

The data for the current study were collected as part of a larger study investigating family relationships and parenting in families of children with FXS. Participants included 23 families of male children with FXS between the ages of 3;0 and 7;11 years, yielding a total of 69 participants including 23 fathers (22 biological fathers and one stepfather), 23 biological mothers, and 23 male children with FXS. See pages 5 – 6 for additional details on the participants in the current study.

In order to address the aims stated above, mothers and fathers independently completed multiple questionnaires via REDCap (Harris et al., 2019; Harris et al., 2009) and one parent completed an interview about the child. Parents completed questionnaires pertaining to: (a) their individual well-being, including the Symptom Checklist-90-Revised (SCL-90-R; Derogatis, 1994) and the Parenting Stress Index – 4th edition, Short Form (PSI-4-SF; Abidin, 2012); (b) couple functioning, including the Couples

Satisfaction Index (CSI-32; Funk & Rogge, 2007) and the Dyadic Coping Inventory (DCI; Bodenmann, 2008; Ledermann et al., 2010); and (c) child functioning and behavior, including the Aberrant Behavior Checklist, 2nd edition (ABC-2; Aman & Singh, 2017) and the Social Responsiveness Scale, 2nd edition SRS-2; Constantino & Gruber, 2012). One parent also completed the Vineland Adaptive Behavior Scales, 3rd edition (Vineland-3; Sparrow, Cicchetti, & Saulnier, 2016) as an interview to assess the child's adaptive behavior.

The SCL-90-R measures mental health symptoms along multiple dimensions yielding a Positive Symptom Total, a Positive Symptom Distress Index, and a Global Severity Index (GSI). T-scores for each dimension and the GSI were reported; the GSI T-score was used in analyses. The PSI-4-SF measures parenting stress, providing subscale scores for Parental Distress, Parent-Child Dysfunctional Interaction, and Difficult Child. T-scores and percentiles from these dimensions as well as the Total Stress score were reported. The Total Stress T-score was used in analyses. The CSI-32 measures couples satisfaction, with higher scores indicating higher levels of relationship satisfaction. Total raw scores were reported and used in analyses. The DCI measures perceived communication and coping that occurs in relationships when one or both partners are experiencing stress. Higher scores indicate higher levels of dyadic coping. Total raw scores from this measure were reported and used in analyses. The ABC-2 measures challenging behaviors of individuals with developmental disabilities in multiple domains. Raw scores from the FXS-specific subscale scoring (Sansone et al., 2012) were reported along with total raw scores; total raw scores were used in analyses. The SRS-2 measures social impairments commonly associated with ASD, providing DSM-5 compatible subscale scores for Social Communication and Interaction (SCI) and Restricted Interests and Repetitive Behavior (RRB), as well as a Total T-score. SCI, RRB, and Total T-scores were reported; Total T-scores were used in analyses. 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 reported and used in analyses. See pages 7 – 10 for additional details regarding the measures used in the current study.

Analysis Plan

All variables were visually inspected to check for model assumptions of normality and homoscedasticity of the residuals. Tests for skewness and kurtosis were also examined. Transformations and nonparametric alternatives were considered for any data that did not meet parametric assumptions.

To address the first and second aims, descriptive summaries of mothers' and fathers' mental health challenges and parenting stress (the outcomes variables) were reported and compared to levels reported in the general population. Then, interspousal correlations were calculated to determine the degree of correspondence between mothers' and fathers' ratings of mental health challenges and parenting stress. Comparisons of mothers' and fathers' mean scores on the SCL-90-R and PSI-4-SF were also reported.

To address the third aim, descriptive summaries of the outcome variables (i.e., couples satisfaction and dyadic coping) were reported and mean scores for mothers and fathers were compared. Interspousal correlations were then calculated to determine the degree of correspondence between mothers' and fathers' ratings of couples satisfaction and dyadic coping. Comparisons of mothers' and fathers' mean scores on the CSI-32 and DCI were also reported. To address the fourth aim, descriptive summaries of the predictor variables (i.e., challenging behaviors, ASD symptoms, and adaptive behavior) and interspousal correlations were reported. Comparisons of mothers' and fathers' mean scores on the ABC-2 and SRS-2 were also reported. Additionally, given that data collected from couples are considered to be non-independent observations (e.g., Hartley, Barker, et al., 2011), a multilevel modeling (MLM) approach, also known as hierarchical linear modeling (HLM), was used for Aims 3 and 4 (Raudenbush & Bryk, 2002). In this approach, the data from each partner is nested within a group that has an *N* of 2 (Campbell & Kashy, 2002). Effect coding was used for parent sex such that Male = 1 and Female = -1. Continuous predictors were centered to their respective grand means.

Visual inspection of the variables and tests for skewness and kurtosis indicated that the CSI-32 scores for mothers and fathers were negatively skewed. A cubic transformation of the variable reduced the negative skewness and was used to examine correlations between the CSI-32 and the other measures.

To avoid difficulty in interpreting the cubic transformation of the CSI-32 variable in a multilevel model, a new categorical variable was created that reduced the significant negative skewness of the CSI-32 scores (confirmed using the Shapiro-Wilk test of normality). For this new variable, ranges of the CSI-32 score were given a value of 1-8 (e.g., scores ≤ 49 had a value of 1, scores from 50 to 99 had a value of 2, scores from 100-109 had a value of 3).

Intraclass correlation coefficients (ICCs) were then calculated to estimate the proportion of the total variation in the dependent variables that exists between versus within couples for Aims 3 and 4. The dependent variables for Aim 3 included couples satisfaction and dyadic coping (total raw scores from the CSI-32 and DCI, respectively). The dependent variables for Aim 4 included couples satisfaction and dyadic coping, as well as mental health challenges (SCL-90-R GSI T-score) and parenting stress (PSI-4-SF Total Stress T-score). Next, multilevel models were specified to examine the outcomes for Aims 3 and 4. For Aim 3, separate models for couples satisfaction and dyadic coping were conducted. The strong and significant association between the variables for parenting stress and mental health challenges did not allow for them both to be included in the models for Aim 3; the parenting stress measure was chosen as it was more strongly associated with both couples satisfaction and dyadic coping than the measure of mental health challenges for both mothers and fathers.

As an example, the model for couples satisfaction (CS) was specified as follows, with parenting stress (PS) and parent sex (sex) set as predictors at Level 1. Covariates included parent age (age) and parent education (edu). In this example, random effects were not included at Level 2 for parenting stress, parent sex, parent age, or parent education; therefore, the effects of these predictors on the outcome (CS) are fixed. However, a family-level random effect for the intercept was included at Level 2:

Level 1:

$$CS_{ij} = \beta_{0j} + \beta_{1j}(PS_{ij}) + \beta_{2j}(sex_{ij}) + \beta_{3j}(age_{ij}) + \beta_{4j}(edu_{ij}) + e_{ij}$$

Level 2:

$$\beta_{0j} = \gamma_{00} + \mu_{0j}$$
$$\beta_{1j} = \gamma_{10}$$
$$\beta_{2j} = \gamma_{20}$$
$$\beta_{3j} = \gamma_{30}$$
$$\beta_{4j} = \gamma_{40}$$

Composite:

$$CS_{ij} = [\gamma_{00} + \gamma_{10}(PS_{ij}) + \gamma_{20}(sex_{ij}) + \gamma_{30}(age_{ij}) + \gamma_{40}(edu_{ij})] + [\mu_{0j} + e_{ij}]$$

For Aim 4, separate models for mental health challenges, parenting stress, couples satisfaction, and dyadic coping were specified. Interactions between parent sex and child variables were included to examine the differential effects of child characteristics on mothers and fathers.

Results

Aims 1 and 2

Aim 1 was to examine mental health challenges and parenting stress in biological mothers of children with FXS, who are carriers of the *FMR1* premutation or the full mutation. Aim 2 was to examine mental health challenges and parenting stress in fathers of children with FXS and to compare maternal and paternal mental health challenges and parenting stress. Table 1 displays descriptive statistics for the SCL-90-R dimension scores, the measure of mental health challenges, for both mothers and fathers. Paired samples t-tests and Wilcoxon signed-ranks tests (when appropriate) confirmed that there were no statistically significant differences between mothers' and fathers' standardized scores on the SCL-90-R.

Table 1 also displays information regarding the number of parents who met the instrument's cutoff for clinical significance on the SCL-90-R dimensions. Overall, a T-score of 63 or above (equivalent to the 90^{th} percentile) on the Global Severity Index, or two or more scores of 63 or above on any dimension, suggest clinically significant levels of mental health challenges. According to these criteria, 10 out of 23 (43%) mothers and 10 out of 23 (43%) fathers in the sample reported clinically significant levels of mental health challenges. Six of these mothers and fathers were from the same family. The rates of clinically significant mental health challenges in this sample are higher than what is reported in the general population for both males and females. Specifically, recent estimates suggest that approximately 24.5% of women in the United States suffer from any mental illness compared to approximately 16.3% of men (SAMHSA, 2019).

Table 1

Mother-Father Comparisons of SCL-90-R Dimension Scores

	Mean T-score (*SD*) Range		*n* (%) with T-score ≥ 63		Interspousal Correlations
SCL-90-R Dimension	Mothers	Fathers	Mothers	Fathers	ρ (*p*-value)
Somatization	50.70 (9.24) 35 – 70	53.57 (12.43) 37 – 77	3 (13%)	4 (17%)	-0.22 (0.320)
Obsessive-Compulsive	58.52 (10.57) 37 – 78	59.39 (12.23) 39 – 80	9 (39%)	9 (39%)	0.14 (0.516)
Interpersonal Sensitivity	57.35 (11.82) 39 – 80	58.04 (13.61) 41 – 80	10 (43%)	9 (39%)	0.03 (0.892)
Depression	57.57 (11.15) 34 – 75	59.22 (13.17) 38 – 80	8 (35%)	9 (39%)	-0.03 (0.877)
Anxiety	52.52 (10.92) 37 – 73	51.65 (11.94) 40 – 73	4 (17%)	5 (22%)	0.11 (0.626)
Hostility	57.30 (8.74) 40 – 74	56.35 (12.46) 41 – 80	6 (26%)	7 (30%)	-0.15 (0.485)
Phobic Anxiety	52.44 (10.76) 44 – 77	52.13 (9.12) 47 – 71	4 (17%)	5 (22%)	0.29 (0.186)
Paranoid Ideation	51.57 (10.48) 41 – 72	55.22 (14.61) 41 – 80	3 (13%)	7 (30%)	0.25 (0.245)
Psychoticism	52.83 (10.56) 44 – 80	51.74 (11.95) 44 – 80	6 (26%)	5 (22%)	0.22 (0.314)
Global Severity Index	56.52 (11.09) 30 – 79	56.78 (14.24) 34 – 80	6 (26%)	8 (35%)	0.02 (0.912)

Table 2 displays descriptive statistics for the PSI-4-SF domain scores, the measure of parenting stress, for both mothers and fathers. Much like the SCL-90-R, paired samples t-tests and Wilcoxon signed-ranks tests (when appropriate) confirmed that there were no statistically significant differences between mothers' and fathers' standardized scores on the PSI-4-SF. Furthermore, on the PSI-4-SF, scores that fall between the 16^{th} and 84^{th} percentiles are considered within the normal range, scores between the 85^{th} and 89^{th} percentiles are considered high, and scores at the 90^{th} percentile and above are within the clinically significant range. Table 3 shows the number of mothers and fathers who reported scores within each of these ranges on the PSI-4-SF domains. Notably, a majority of mothers and fathers reported normal levels of parenting stress on the Parental Distress and Parent-Child Dysfunctional Interaction domains. However, a fairly large proportion of both mothers (43%) and fathers (30%) reported clinically significant levels of parenting stress in the Difficult Child domain.

Table 2

Mother-Father Comparisons of PSI-4-SF Domain Scores

	Mothers		Fathers		Interspousal Correlation
	Mean (*SD*) Range		Mean (*SD*) Range		
PSI-4-SF Domain	T-Score	Percentile	T-Score	Percentile	ρ (*p*-value)
Parental Distress	54.09 (12.41) 34 – 79	61.39 (30.57) 3 – 99	50.74 (10.30) 34 – 72	54.30 (28.22) 3 – 99	0.21 (0.348)
P-C Dysfunctional Interaction	55.30 (8.55) 41 – 76	68.78 (19.97) 24 – 99	55.35 (9.19) 40 – 81	68.39 (18.55) 19 – 99	0.04 (0.849)
Difficult Child	59.91 (9.34) 42 – 80	78.09 (19.51) 26 – 99	57.70 (10.56) 35 – 77	72.26 (24.89) 6 – 99	0.30 (0.170)
Total Stress	57.04 (10.24) 41 – 81	69.96 (23.21) 19 – 99	54.87 (9.73) 35 – 79	66.09 (23.28) 4 – 99	0.23 (0.288)

To determine the degree of correspondence between mothers' and fathers' ratings of mental health challenges and parenting stress, interspousal correlations were calculated. Table 1 and Table 2 also display interspousal correlations for the SCL-90-R dimensions and PSI-4-SF domains, respectively. Given that some of the scores for these two measures were not normally distributed, Spearman's rank-order correlations were reported instead of Pearson's correlations. Surprisingly, no significant correlations were found between mothers' and fathers' scores across these measures despite the overlap within families in clinically significant cases on the SCL-90-R and the fact that there were no significant differences in the mean scores between mothers and fathers on either of the measures.

Table 3

PSI-4-SF Domain Percentile Scores

	Percentile Range *n*, (%)					
	Normal (16 – 84)		High (85 – 89)		Clinically Significant (90+)	
PSI-4-SF Domain	Mothers	Fathers	Mothers	Fathers	Mothers	Fathers
Parental Distress	17 (74%)	21 (91%)	1 (4%)	0 (0%)	5 (22%)	2 (9%)
P-C Dysfunctional Interaction	18 (78%)	20 (87%)	1 (4%)	1 (4%)	4 (17%)	2 (9%)
Difficult Child	12 (52%)	15 (65%)	1 (4%)	1 (4%)	10 (43%)	7 (30%)
Total Stress	17 (74%)	18 (78%)	0 (0%)	2 (9%)	6 (26%)	3 (13%)

Note. The individual percentage values are rounded and may not total 100%. Parents whose percentile scores were below 16 were included in the Normal category.

Aim 3

Aim 3 was to examine relationships between aspects of the couple relationship (i.e., couples satisfaction and dyadic coping) and mothers' and fathers' mental health challenges and parenting stress. The ICC for couples satisfaction indicated that 76.6% of the variation was due to between-couples factors whereas 23.4% was due to within-couple factors. For dyadic coping, 42.1% of the variation was due to between-couples factors whereas 57.9% was due to within-couple factors. Table 4 displays descriptive statistics for mothers' and fathers' CSI and DCI scores as well as interspousal correlations, which indicate the degree of correspondence between their ratings of couples satisfaction and dyadic coping.

Unlike the measures of mental health challenges and parenting stress, interspousal correlations indicated that there were significant correspondences between mothers' and fathers' scores on both the CSI-32 and DCI, with mean scores indicating average levels of couples satisfaction and dyadic coping for both mothers and fathers. Additionally, paired samples t-tests confirmed that there were no statistically significant differences between mothers' and fathers' scores on the CSI-32 or the DCI. Overall, only six mothers (26%) and four fathers (17%) reported notable relationship dissatisfaction on the CSI-32.

Additionally, on the DCI, five mothers and five fathers (22%) reported below average levels of dyadic coping, 13 mothers (57%) and 14 fathers (61%) reported average levels of dyadic coping, and five mothers (22%) and four fathers (17%) reported above average levels of dyadic coping.

Table 4

Mother-Father Comparisons of Couple Relationship Measures

	Mean (*SD*) Range		*r* (*p*-value)
Variable	Mothers	Fathers	Interspousal Correlation
Couples Satisfaction[1]	123.74 (30.28) 33 – 155	124.57 (30.82) 43 – 158	**0.76***** **(0.0001)**
Dyadic Coping	130.78 (18.74) 98 – 167	125.83 (16.26) 100 – 156	**0.44*** **(0.038)**

Note. [1]CSI-32 raw score underwent cubic transformation to reduce negative skewness.
* $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$.

Prediction of Couples Satisfaction

Table 5 presents the results of the MLM analysis for couples satisfaction. As expected, there was a significant main effect of parenting stress on couples satisfaction. Across all couples, when the other predictors were at their mean values, parents with greater than average parenting stress experienced less couples satisfaction than parents with less than average parenting stress. There were no significant main effects of parent sex, parent age, or parent education on couples satisfaction.

Prediction of Dyadic Coping

Table 5 also presents the results of the MLM analysis for dyadic coping. As predicted, there was a significant main effect of parenting stress on dyadic coping. Across all couples, when the other predictors were at their mean values, parents with greater than average parenting stress experienced less dyadic coping than parents with less than average parenting stress. There was also a main effect of parent education on dyadic coping. Across all couples, when the other predictors were at their mean values, parents with greater than average levels of education experienced less dyadic coping than parents with less than average levels of education. There was no significant main effect of parent sex on dyadic coping, but there was a marginally significant main effect of parent age on dyadic coping ($p = 0.083$), suggesting that across all couples, when the other predictors were at their mean values, older parents experienced less dyadic coping than younger parents.

Table 5

Multilevel Model Results for Aim 3

	Couples Satisfaction	Dyadic Coping
Fixed Effects		
Intercept	5.30***	128.30***
	(0.36)	(2.32)
Parenting Stress	-0.07*	-0.94***
	(0.03)	(0.23)
Parent Sex	0.01	-2.16
	(0.20)	(1.99)
Parent Age	-0.10	-0.71~
	(0.06)	(0.41)
Parent Education	-0.21	-3.47*
	(0.19)	(1.61)
Random Effects		
Residual (σ_e^2)	1.72	169.75
Intercept (σ_{u0}^2)	2.10	39.30
Goodness-of-fit		
AIC	205.33	376.16
BIC	218.13	388.96

Note. Standard errors in parentheses.
~ $p < 0.10$, * $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$.

Aim 4

Aim 4 was to examine the contributions of child challenging behaviors, ASD symptoms, and adaptive behavior to parental individual well-being (i.e., mental health challenges and parenting stress) and couple well-being (i.e., couples satisfaction and dyadic coping). The ICC for mental health challenges indicated that 3.1% of the variation was due to between-couples factors whereas 96.9% was due to within-couple factors. For parenting stress, 27.0% of the variation was due to between-couples factors whereas 73.0% was due to within-couple factors. Visual inspection of the variables and tests for kurtosis and skewness indicated that several of the ABC-2 subscale scores and one of the SRS-2 subscale scores were not normally distributed. Tables 6 and 7 display descriptive statistics for parents' ABC-2 and SRS-2 scores, respectively, as well as interspousal correlations, which indicate the degree of correspondence between their ratings of child challenging behaviors and ASD symptoms on these measures. Given that some of the scores for these two measures 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 mothers' and fathers' scores on the ABC-2 for the Hyperactivity and Inappropriate Speech subscales on the ABC-2, but not for the other four subscales. On the SRS-2, there were significant correspondences between mothers' and fathers' scores on the SCI and RRB subscale T-scores as well as the Total T-score. Additionally, paired samples t-tests and Wilcoxon signed-ranks tests (when appropriate) confirmed that there were no significant differences between mothers' and fathers' subscale and total scores on these measures except for the ABC-2 Hyperactivity subscale, $t(22) = 2.11$, $p = 0.046$, and the SRS-2 RRB subscale, $Z = 2.27$, $p = 0.023$. For both of these subscales, mothers endorsed higher scores than fathers.

Table 6

Mother-Father Comparisons of ABC-2 Subscale Raw Scores

	Mean (*SD*) Range		ρ (*p*-value)
ABC-2 Subscale	Mothers	Fathers	Interspousal Correlation
Irritability	18.26 (10.62) 1 – 38	17.44 (11.54) 1 – 47	0.36 (0.096)
Socially Unresponsive / Lethargic	4.52 (5.86) 0 – 23	5.17 (4.65) 0 – 20	0.23 (0.299)
Stereotypy	6.09 (2.78) 1 – 12	4.96 (3.76) 0 – 13	0.24 (0.268)
Hyperactivity	14.04 (8.07) 2 – 27	11.00 (5.42) 1 – 24	**0.54**** **(0.007)**
Inappropriate Speech	4.48 (3.85) 0 – 11	3.17 (2.29) 0 – 8	**0.48*** **(0.021)**
Social Avoidance	1.39 (2.61) 0 – 9	1.52 (2.39) 0 – 9	0.33 (0.123)
Total Score	49.39 (27.71) 12 – 98	43.87 (24.70) 10 – 103	0.36 (0.094)

Note. * $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$.

Furthermore, according to the SRS-2 Total Score guidelines, scores can be classified as within normal limits (T-scores ≤ 59), in the mild range (T-scores = 60 to 65), in the moderate range (T-scores = 66 to 75), or in the severe range (T-scores ≥ 76). Table 8 displays the number of mothers and fathers who reported scores within each of these ranges on the SRS-2. Additionally, Table 9 displays descriptive statistics for the Vineland-3 scores, and Tables 10 and 11 display correlations between the measures of couple (i.e., CSI-32 and DCI total raw scores), individual (i.e., SCL-90-R Global Severity Index and PSI-4-SF Total Stress T-scores), and child (ABC-2 total raw score and SRS-2 Total T-Score) functioning for mothers and fathers, respectively.

Table 7

Mother-Father Comparisons of SRS-2 Subscale T-Scores

	Mean (*SD*) Range		ρ (*p*-value)
SRS-2 Subscale	Mothers	Fathers	Interspousal Correlation
Social Communication and Interaction	69.00 (11.00) 49 – 90	66.83 (9.53) 49 – 85	**0.56**** **(0.005)**
Restricted Interests and Repetitive Behavior	75.48 (11.63) 57 – 92	69.17 (10.35) 54 – 90	**0.41*** **(0.049)**
Total Score	70.87 (10.96) 51 – 91	67.57 (9.40) 51 – 87	**0.52*** **(0.011)**

Note. * $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$.

Table 8

Mother-Father Comparison of SRS-2 Total Score Severity Range

	n, (%)	
Range	Mothers	Fathers
Within normal limits	3 (13%)	5 (22%)
Mild	4 (17%)	4 (17%)
Moderate	9 (39%)	8 (35%)
Severe	7 (30%)	6 (26%)

Note. The individual percentage values are rounded and may not total 100%.

Table 9

Vineland-3 Scores

Vineland-3 Domain	Mean (*SD*)	Range
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

The significant correlations between the ABC-2 and SRS-2 scores for both mothers and fathers (see Tables 10 and 11) indicated that they should not be included together in the MLMs for Aim 4. There was also a significant correlation between mothers' SRS-2 scores and the Vineland-3 Adaptive Behavior Composite (r = -0.72, p = 0.001) and a marginally significant correlation between fathers' SRS-2 scores and the Vineland-3 Adaptive Behavior Composite (r = -0.36, p = 0.085). Therefore, the ABC-2 Total Score, Vineland-3 Adaptive Behavior Composite, child age, and parent sex were included as predictors in the MLMs for Aim 4, as well as an interaction between the ABC-2 Total Score and parent sex. Table 12 presents the results of the MLM analyses for mental health challenges, parenting stress, couples satisfaction, and dyadic coping.

Table 10

Pearson Correlations for Maternal Variables

Variable	1.	2.	3.	4.	5.	6.
1. SCL-90-R[1]	1.00					
2. PSI-4-SF[2]	**0.73***** **(0.0001)**	1.00				
3. CSI-32[3]	-0.29 (0.181)	**-0.44*** **(0.037)**	1.00			
4. DCI[4]	-0.27 (0.212)	**-0.53*** **(0.010)**	**0.81***** **(0.0001)**	1.00		
5. ABC-2[5]	**0.65**** **(0.001)**	**0.64**** **(0.001)**	**-0.55**** **(0.006)**	**-0.54**** **(0.007)**	1.00	
6. SRS-2[6]	0.29 (0.188)	0.32 (0.139)	**-0.50*** **(0.016)**	-0.35 (0.098)	**0.47*** **(0.026)**	1.00

Note. [1]Global Severity Index T-score used for SCL-90-R. [2]Total Stress T-score used for PSI-4-SF. [3]CSI-32 raw score underwent cubic transformation to reduce negative skewness. [4]DCI raw score. [5]ABC-2 Total raw score. [6]SRS-2 Total T-score.
* $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$.

Table 11

Pearson Correlations for Paternal Variables

Variable	1.	2.	3.	4.	5.	6.
1. SCL-90-R[1]	1.00					
2. PSI-4-SF[2]	**0.64**** **(0.001)**	1.00				
3. CSI-32[3]	-0.25 (0.243)	**-0.68***** **(0.0004)**	1.00			
4. DCI[4]	-0.40 (0.058)	**-0.47*** **(0.022)**	**0.52*** **(0.011)**	1.00		
5. ABC-2[5]	0.37 (0.082)	**0.59**** **(0.003)**	**-0.46*** **(0.028)**	-0.39 (0.063)	1.00	
6. SRS-2[6]	**0.55**** **(0.007)**	**0.62**** **(0.001)**	**-0.54**** **(0.008)**	-0.27 (0.216)	**0.72***** **(0.0001)**	1.00

Note. [1]Global Severity Index T-score used for SCL-90-R. [2]Total Stress T-score used for PSI-4-SF. [3]CSI-32 raw score underwent cubic transformation to reduce negative skewness. [4]DCI raw score. [5]ABC-2 Total raw score. [6]SRS-2 Total T-score.
* $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$.

Prediction of Parent Mental Health Challenges

As expected, there was a significant main effect of child challenging behaviors on parental mental health challenges. Across all couples, when the other predictors were at their mean values, parents of children with greater than average levels of challenging behaviors experienced more mental health challenges than parents of children with less than average levels of challenging behaviors. However, there were no significant main effects of child adaptive behavior, child age, or parent sex on parent mental health challenges, nor was there a significant interaction between child challenging behaviors and parent sex in predicting mental health challenges. Model diagnostics suggested that a linear regression model would be sufficient for predicting mental health challenges. The results of a linear regression model were similar to the results of the multilevel model.

Prediction of Parenting Stress

As expected, there was a significant main effect of child challenging behaviors on parenting stress. Across all couples, when the other predictors were at their mean values, parents of children with greater than average levels of challenging behaviors experienced more parenting stress than parents of children with less than average levels of challenging behaviors. There was also a significant main effect of child adaptive behavior on parenting stress, suggesting that across all couples, when the other predictors were at their mean values, parents of children with greater than average levels of adaptive behavior experienced less parenting stress than parents of children with less than average levels of adaptive behavior. There were no significant main effects of child age or parent sex on parenting stress, nor was there a significant interaction between child challenging behaviors and parent sex in predicting parenting stress. Model diagnostics suggested that a linear regression model would also be sufficient for predicting parenting stress. The results of a linear regression model were similar to the results of the multilevel model.

Table 12

Multilevel Model Results for Aim 4

	Mental Health Challenges	Parenting Stress	Couples Satisfaction	Dyadic Coping
Fixed Effects				
Intercept	56.58^{***}	55.91^{***}	5.29^{***}	128.51^{***}
	(1.73)	(1.16)	(0.32)	(2.68)
Challenging Behavior	0.23^{**}	0.21^{***}	-0.03^{**}	-0.27^{**}
	(0.07)	(0.05)	(0.01)	(0.10)
Adaptive Behavior	0.001	-0.21^{*}	0.07^{*}	0.21
	(0.15)	(0.10)	(0.03)	(0.25)
Child Age	-0.50	-0.89	-0.34	0.55
	(1.28)	(0.86)	(0.23)	(2.04)
Parent Sex	0.77	-0.51	-0.13	$-3.22^{\sim}$
	(1.72)	(1.16)	(0.18)	(1.90)
Challenging Behavior x Parent Sex	-0.03	-0.02	-0.01	0.08
	(0.06)	(0.05)	(0.01)	(0.08)
Random Effects				
Residual (σ^2_e)	134.34	61.60	1.42	163.09
Intercept (σ^2_{u0})	0.89	1.29e-17	1.63	94.18
Goodness-of-fit				
AIC	367.19	335.75	207.95	389.43
BIC	381.82	350.38	222.58	404.06

Note. Standard errors in parentheses.
$^{\sim}$ $p < 0.10$, * $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$.

Prediction of Couples Satisfaction

As expected, there was a significant main effect of child challenging behaviors on couples satisfaction. Across all couples, when the other predictors were at their mean values, parents of children with greater than average levels of challenging behaviors experienced less couples satisfaction than parents of children with less than average levels of challenging behaviors. There was also a significant main effect of child adaptive behavior on couples satisfaction. Across all couples, when the other predictors were at their mean values, parents of children with greater than average levels of adaptive behavior experienced more couples satisfaction than parents of children with less than average levels of adaptive behavior. There were no significant main effects of child age or parent sex on couples satisfaction, nor was there a significant interaction between child challenging behaviors and parent sex in predicting couples satisfaction.

Prediction of Dyadic Coping

As expected, there was a significant main effect of child challenging behaviors on dyadic coping. Across all couples, when the other predictors were at their mean values, parents of children with greater than average levels of challenging behaviors experienced lower levels of dyadic coping than parents of children with less than average levels of challenging behaviors. There was also a marginally significant main effect of parent sex on dyadic coping ($p = 0.091$). In reference to the overall mean, when the other predictors were at their mean values, fathers reported lower levels of dyadic coping compared to mothers. There were no significant main effects of child adaptive behavior or child age on dyadic coping, nor was there a significant interaction between child challenging behaviors and parent sex in predicting dyadic coping.

Discussion

The current study was designed to examine aspects of the broader family environment in families of young children with FXS, including maternal and paternal well-being, features of the mother-father relationship, and relationships between child characteristics and parent and couple functioning. Findings suggest that both mothers and fathers of young children with FXS are at risk for experiencing significant mental health challenges and parenting stress. Unfortunately, nearly half of all mothers and fathers reported clinically significant levels of mental health challenges on the SCL-90-R. These rates are notably higher compared to what is observed in the general population for both males and females (SAMHSA, 2019). An abundance of past research has established that women with the *FMR1* premutation experience mental health problems independent of the stress associated with parenting a child or multiple children with significant challenges (Hagerman et al., 2018). The results of the current study confirm that fathers in these families are also experiencing substantial mental health problems and that both parents may be in need of greater support and services.

Additionally, 43% of mothers and 30% of fathers reported clinically significant levels of parenting stress in the Difficult Child domain on the PSI-4-SF. Parents reported greater stress in the Difficult Child domain compared to the other domains (i.e., Parental Distress and Parent-Child Dysfunctional Interaction), suggesting that their perceptions of the child's behavior were contributing more to their stress than their adjustment to parenting or their relationship with their child. This profile of parenting stress is consistent with past research on mothers of children with FXS (Johnston et al., 2003; Wheeler et al., 2007). Interestingly, there were no significant correlations between parents' scores on the SCL-90-R or the PSI-4-SF. The lack of associations between parents' scores on these measures potentially suggests that within families, one parent may be compensating for or supporting a partner who is struggling with mental health or parenting stress, potentially buffering against negative effects on the child. However, on the SCL-90-R, there was overlap for six families such that both the mother and father reported clinically significant levels of mental health challenges. Therefore, developing a better understanding of these dynamics within families is an area for future research. Additionally, an important consideration regarding these findings is that the majority of these data were collected during the COVID-19 pandemic, which may have contributed to parents' mental health challenges and stress related to parenting, especially given that services for children with developmental disabilities were severely interrupted during this time (Abbeduto, 2020; Chan & Fung, 2021; Constantino et al., 2020; Manning et al., 2020; White et al., 2021).

Despite experiencing challenges with mental health and parenting stress, most mothers and fathers reported moderate to high levels of couples satisfaction and dyadic coping, with very few parents reporting notable relationship dissatisfaction, and a majority of parents reporting dyadic coping in the average or above average range. In these families, higher levels of couples satisfaction and dyadic coping may be protective against the daily stressors that the parents are experiencing (McCarthy et al., 2006). Importantly, features of the mother-father relationship are likely to affect the mother-child and father-child relationships. Specifically, parents with higher levels of couples satisfaction and dyadic coping may be more likely to engage in positive and responsive interactions with their children (e.g., Belsky, 1981; Belsky, 1984; Kouros et al., 2014; Peltz et al., 2018). However, one limitation of these results is the possibility of selection bias such that only relatively satisfied couples were willing to participate in the current study.

Mothers and fathers also reported independently on their child's challenging behaviors and ASD symptoms. Interspousal correlations indicated high degrees of correspondence between mothers' and fathers' scores on the SRS-2, but not the ABC-2. On average, mothers and fathers reported moderate levels of challenging behaviors that were similar to the ABC scores reported in Sansone et al. (2012). With regard to ASD symptoms, a majority of both mothers and fathers reported scores that fell within the moderate to severe range, indicating that many of the children in this sample were demonstrating deficiencies in reciprocal social behavior that may interfere with everyday social interactions. Although significant correspondences were found between mothers' and fathers' scores on the SRS-2, mothers on average reported higher levels of behaviors in the RRB subscale compared to fathers. Mothers may be more likely to observe these behaviors, especially given that 16 of the 23 families in the study reported that the mother spent more time with the child compared to the father. This difference in time spent with the child may also influence the lack of correspondences between mothers' and fathers' scores on the ABC-2. Fathers may be observing fewer of the child's challenging behaviors when the mother is the child's primary caregiver.

There were also some interesting differences in the correlations between maternal and paternal measures. For mothers (but not fathers), there were strong and significant correlations between the ABC-2 and the other measures of individual, couple, and child functioning (i.e., the SCL-90-R GSI score, the PSI-4-SF Total Stress T-Score, the CSI-32 raw score, the DCI raw score, and the SRS-2 Total T-score). However, for fathers (but not mothers), the SRS-2 was strongly correlated with every measure except the DCI. This finding may be due to differences in parental experiences of challenging behaviors and ASD symptoms; that is, mothers may be experiencing and managing more challenging behaviors compared to fathers, and fathers may be more concerned about or influenced by the child's ASD symptoms compared to mothers. In particular, paternal parenting stress was associated with the child's ASD symptoms, whereas maternal parenting stress was not. However, consistent with past research, parenting stress for both mothers and fathers was related to child challenging behaviors (e.g., Johnston et al., 2003; McCarthy et al., 2006). Future studies should investigate the similarities and differences between mothers and fathers further to determine how parents' impressions of the child's behavior influence their well-being.

Additionally, parenting stress was found to associate with both couples satisfaction and dyadic coping, with no significant differences found between mothers and fathers. Child challenging behavior was also found to associate with parental mental health challenges, parenting stress, couples satisfaction, and dyadic coping. Surprisingly, no significant differences were found between mothers and fathers across these analyses, including any differences between mothers and fathers based on child challenging behaviors. Perhaps future investigations with larger sample sizes would find differences between parents. Child adaptive behavior was also found to associate with couples satisfaction and parenting stress. These findings emphasize the importance of early intervention for children with FXS focused not only on communication and socialization skills, but also daily living skills that promote independence.

Interventions focused on reducing parenting stress in these families could also have a positive impact on parents' individual well-being and the mother-father relationship. One potential intervention that could be beneficial for parents of children with FXS is Mindfulness-Based Stress Reduction (MBSR). MBSR is an established and empirically supported stress-reduction intervention that has been shown to reduce parental stress, depressive symptoms, and parent-reported child behavior problems in families of children with developmental disabilities (e.g., Chan & Neece, 2018; Neece et al., 2019). Another study of MBSR for parents of children with developmental disabilities also found improvements in child social skills that were mediated by parent-child relational factors (i.e., attachment and discipline practices; Lewallen & Neece, 2015). Interestingly, a recent study of mothers of children with FXS found that trait mindfulness, acceptance, and mindful parenting were associated with lower levels of anxiety, depression, and stress (Wheeler et al., 2018), providing additional evidence that mindfulness interventions may be beneficial for these families. Based on the findings of the current study, reductions in parental stress are likely to benefit parental individual well-being and couple well-being with anticipated benefits for the parent-child relationship as well.

Parent-implemented interventions focused on teaching parents strategies for managing child challenging behaviors and engaging in responsive interactions may also benefit parental and couple well-being in families of children with FXS. A recent study by Hall et al. (2020) examined the effects of functional communication training (FCT) delivered via telehealth on problem behaviors in young boys with FXS. Children with FXS often engage in problem behaviors that serve different communicative functions, including gaining access to attention or a highly preferred item or escaping a demanding task or situation. The focus of FCT is to ensure that these problem behaviors are no longer reinforced by the caregiver while simultaneously teaching the child alternative and appropriate ways to communicate their preferences and needs. The FCT intervention conducted by Hall and colleagues (2020) led to significant reductions in child problem behaviors as well as decreased levels of parenting stress, likely benefiting the entire family system.

The recent FCT study, along with other parent-implemented intervention studies conducted in the past several years with families of children with FXS (e.g., McDuffie et al., 2018; Thurman et al., 2020), support the use of telehealth as an effective service delivery model for families of children with FXS. The use of telehealth-enabled interventions in this relatively rare population also allows families from rural and/or underserved communities to participate in research studies and receive services that may otherwise not be available to them (Abbeduto, 2020; Abbeduto et al. 2019; Hall et al., 2020). Telehealth also offers more flexibility compared to in-person services and is more cost-effective (Abbeduto, 2020; McDuffie et al., 2016). Therefore, telehealth methods for conducting assessments with and delivering interventions to families affected by FXS are likely to be utilized more frequently in the post-pandemic world.

Limitations and Future Directions

There are some notable limitations to this study, including the relatively small sample size and the lack of diversity in the sample. FXS research studies focused on parent-child relationships tend to have small samples and the majority of the sample is typically families who identify as white, highly educated, and have household incomes in the middle to high range. Therefore, future studies should attempt to reduce barriers to participation in research for FXS families from underrepresented groups. These barriers include age of diagnosis, lack of information about research opportunities, time commitment for participation in research, and low household income (Chechi et al., 2014; Visootsak et al., 2011). Future research should identify ways to reduce these barriers and extend outreach to groups underrepresented in FXS research.

One notable strength of the study is the inclusion of both mothers and fathers as independent informants given that the majority of past research in FXS has focused on the mother-child dyad. Fathers have been historically underrepresented in research on child and adolescent development, both in the general population and in families that include children with disabilities, despite the fact that fathers have a unique and independent role in parenting compared to mothers and may differentially affect the child's development (Cabrera et al., 2018; Fabiano & Caserta, 2018; Phares et al., 2005). For decades, scholars have recognized the importance of the father's role in the family and made suggestions for future research that involve conceptualizing the family as a complex and dynamic system (Cabrera et al., 2014; Cabrera et al., 2000; Phares, 1992), but very little progress has been made in this regard, particularly as it concerns families that include a child with a developmental disability (Braunstein et al., 2013; Hartley, Seltzer, Head, & Abbeduto, 2012). Future studies in these families should continue to include fathers, and also consider differences between two-parent families and single-parent families. These approaches will increase understanding of the family as a complex and dynamic system that differentially influences the development of each family member.

Additionally, parents provided the measures of child ASD symptoms and challenging behaviors as opposed to these behaviors being rated by an independent informant. Future studies should incorporate multiple distinct assessments of both parent and child functioning to ensure accurate measurement within various domains of behavioral and psychological functioning. Furthermore, biological markers of stress were not collected nor were any measures of IQ. Future studies could benefit from including these variables. Another limitation was the focus on concurrent associations as opposed to longitudinal ones. Future studies should examine relationships between parent and child functioning over time to develop a better understanding of how these relationships fluctuate as the child develops. Finally, given that the majority of families participated in the study during the COVID-19 pandemic, the data reported in the current study may not reflect typical family functioning in families of children with FXS.

Conclusions

The findings from the current study indicate the importance of considering the entire family system in families affected by FXS. Both mothers and fathers are in need of greater support to reduce their mental health challenges and parenting stress, which would likely benefit not only parental well-being, but also the mother-father relationship and the relationships between each parent and the child. These results also provide evidence that child challenging behaviors and limited adaptive functioning influence the mother-father relationship as well as individual parent functioning. Early intervention for children with FXS, parent-implemented interventions focused on managing challenging behaviors, and parent interventions focused on reducing stress are likely to benefit these families. Future studies should continue to investigate the complex dynamics between mothers, fathers, and children in families affected by FXS.

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Study 2: The Influence of Parent and Couple Characteristics on Parental Responsivity During Parent-Child Interactions in Families of Children with Fragile X Syndrome

Abstract

Parents of children with fragile X syndrome (FXS) experience elevated levels of parenting stress due to the challenges associated with raising a child with significant impairments. Moreover, biological mothers of children with FXS are at an increased genetic risk for experiencing mental health challenges, including anxiety and depression. Parental mental health challenges and stress are often associated with reduced marital cohesion and satisfaction, which is likely to spill over and negatively affect the parent-child relationship for both mothers and fathers. The current study was designed to examine relationships among characteristics of parents and couples and parent behavior (i.e., responsivity and behavior management) during mother-child and father-child dyadic interactions in 23 families of young boys with FXS. Results indicate that mothers and fathers used similar rates of responsive behaviors, but that fathers used higher rates of behavior management strategies compared to mothers. Parenting stress predicted lower rates of parental responsivity and higher rates of behavior management, but these effects were only marginally significant. Couples satisfaction was not found to associate with either parental responsivity or behavior management, despite the significant relationship between parenting stress and couples satisfaction. Overall, this study provides evidence that reducing parenting stress may lead to more responsive parent-child interactions, and equally so for both mothers and fathers. Therefore, family-based services for families of children with FXS should include interventions that specifically target the elevated rates of parenting stress in these families.

Keywords: fragile X syndrome, parenting stress, couples satisfaction, parental responsivity, behavior management

Many of the conditions and symptoms associated with fragile X syndrome (FXS), including intellectual disability (ID), autism spectrum disorder (ASD), anxiety, and challenging behaviors, can make it difficult for parents to engage in the sustained, productive, and responsive interactions with their children that support optimal development (e.g., McDuffie et al., 2018). Mothers of children with FXS may also have greater difficulty compared to mothers of neurotypical children or children with other developmental disabilities maintaining high levels of responsivity given the physical and mental health challenges associated with being a carrier of the *FMR1* premutation (Hagerman et al., 2018). Therefore, it is important to identify the ways in which mothers of children with FXS can support their child's language learning from an early age.

Fathers have been historically underrepresented in developmental research, including research on FXS, despite the fact that they play a unique and complementary role in parenting compared to mothers (Cabrera et al., 2018). Past research has shown that high levels of father involvement are associated with positive cognitive, social, and behavioral outcomes in neurotypical children (Keown et al., 2018; Wilson & Prior, 2011). In contrast, paternal depression has been linked to less time spent interacting with the child and engaging in supportive activities such as shared book reading, with negative downstream effects on child language (Paulson et al., 2009), as well as negative social and behavioral outcomes (Gross et al., 2008). Unfortunately, fathers of children with FXS also experience elevated levels of parenting stress (McCarthy et al., 2006), which reduces the likelihood for highly positive and responsive father-child interactions (Ward & Lee, 2020).

Additionally, dysfunction or problems in the marital relationship may negatively affect the parent-child relationship for both mothers and fathers (England & Sim, 2009). Family systems theory (Cox & Paley, 1997) emphasizes the potential value in expanding the focus beyond the dyad to consider the ways in which broader family processes and relationships might influence both dyadic and individual development and vice versa. Consequently, features of dyadic subsystems in the family (i.e., the mother-father dyad, mother-child dyad, and father-child dyad) need to be examined in families of children with FXS in order to gain a better understanding of how the family environment influences dyadic and individual functioning across multiple domains. For example, according to the “spillover” hypothesis, mothers and fathers who have stable and healthy relationships are more likely to have positive and responsive parent-child relationships and thereby, children with more optimal outcomes (e.g., Belsky, 1981; Belsky, 1984; Greenlee et al., 2021; Kouros et al., 2014; Peltz et al., 2018). The current study examined the ways in which features of mothers and fathers, as well as features of the mother-father relationship, influence parents’ use of responsive behaviors during dyadic interactions with their child.

Parental Responsivity

Parental responsivity has been defined in past FXS research as, “a healthy, growth-producing relationship consisting of such caregiver characteristics as warmth, nurturance, stability, predictability, and contingent responsiveness” (Spiker et al., 2002, p. 37). Parental responsivity is typically coded from video observations of a parent and child interacting in one or more contexts, including at home or in a clinic setting, at either a molecular or molar level (Warren & Brady, 2007). These contexts include shared book reading, preparing and eating a snack together, free play, and various daily living activities. *Molecular* analyses of responsivity entail the coding of parental behaviors on a behavior-by-behavior basis, whereas *molar* responsivity entails the use of a rating scale to capture the rater’s global impressions of parental affect and parenting style. Molecular-level parental responsivity includes behaviors such as commenting on the child’s focus of attention and asking questions that encourage the child to participate in the interaction. The more frequent use of these parent behaviors is related to positive cognitive, language, and social outcomes for neurotypical children (Landry et al., 2006; Landry et al., 2001; Rowe, 2012; Tamis-LeMonda et al., 2001; Tamis-LeMonda et al., 2014), as well as for children with developmental disabilities, including Down syndrome (DS), ASD, and FXS (McDuffie & Yoder, 2010; Sterling & Warren, 2014; Warren et al., 2010; Yoder & Warren, 1998, 1999, 2001, 2004). The current study examined molecular-level responsivity in both mother-child and father-child play-based interactions in families of children with FXS.

To date, only a small body of work has examined the effects of parental responsivity on child language in FXS as well as factors that contribute to variability in responsivity. In the last decade, Warren and colleagues have published a number of studies on a cohort of 55 mother-child dyads. These studies have demonstrated that both early and sustained maternal responsivity are critically important for child outcomes across a variety of domains (Brady et al., 2020; Brady et al., 2014; Warren, et al., 2017; Warren et al., 2010). Specifically, maternal responsivity predicted receptive and expressive language outcomes in early childhood (Warren et al. 2010), middle childhood (Brady et al., 2014), and adolescence (Brady et al., 2020) in youth with FXS. Moreover, maternal responsivity predicted adaptive behavior outcomes in middle childhood in the domains of Communication, Socialization, and Daily Living Skills (Warren et al., 2017). This research has also demonstrated that wide range of both maternal and child factors are associated with variability in maternal behavior, including maternal education and IQ, maternal depression, child developmental level, and rate of child communication (Sterling et al., 2013).

Studies have also shown that mothers of children with FXS are able to successfully adapt to their children in supportive and responsive ways despite the phenotypic features associated with both the child's FXS and the mother's status as a carrier of the *FMR1* premutation (Sterling et al., 2012; Sterling & Warren, 2018; Sterling et al., 2013). Additionally, recent studies of parent-implemented language interventions by McDuffie, Abbeduto, and colleagues found that parents are able to successfully implement targeted responsive strategies that they are taught to use and that there are associated gains in child engagement and language as a result (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; Thurman et al., 2020). Interestingly, across these studies, there was a total of 61 mother-child dyads and only one father-child dyad.

Fathers of Children with Fragile X Syndrome

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).

Factors Related to Variability in Parental Responsivity

There are multiple parental and family-related factors that contribute to variability in parental responsivity, yet the majority of research, whether involving children with FXS, other developmental disorders, or neurotypical development, has focused exclusively on the mother-child dyad. Past studies of families of children with FXS have shown that maternal depressive symptoms and stress, as well as maternal IQ and education, relate to variability in maternal responsivity (Sterling et al., 2013; Wheeler et al., 2007). Additionally, a study of mother-child dyads in families of children with FXS found associations between maternal physiological arousal (measured through salivary cortisol) and maternal responsivity toward the child with FXS (Robinson et al., 2016). Moreover, in families of young children with DS, parental depression was found to be associated with lower levels of expressive language development (D'Souza et al., 2020), with studies of neurotypical children linking parental depression to reduced parental responsivity and associated negative effects on child outcomes (e.g., Justice et al., 2019; Paulson et al., 2009). Another study of children with ID of heterogenous etiology found that maternal anxiety and depression were negatively related to child language outcomes, and that maternal responsiveness and paternal teaching were positively related to child cognitive and language outcomes (Vilaseca et al., 2019). Aspects of the mother-father relationship, including couples satisfaction, also influence each parents' behavior and relationship with their child (e.g., Fink et al., 2020; Pratt et al., 1992; Peltz et al., 2018; Stroud et al., 2011), but these associations have not yet been explored in families of children with FXS.

Current study

The current study was designed to examine relationships among parental and couple characteristics and parental responsivity in mother-child and father-child dyadic interactions. The first aim was to examine relationships among maternal responsivity, paternal responsivity, and parent individual well-being (i.e., mental health challenges and parenting stress). We hypothesized that mothers and fathers who endorsed higher levels of individual well-being (i.e., lower levels of mental health challenges and parenting stress) would demonstrate higher rates of parental responsivity in the dyadic parent-child interactions (Sterling et al., 2013). The second aim was to examine relationships among maternal responsivity, paternal responsivity, and couple well-being (i.e., couples satisfaction and dyadic coping). We hypothesized that mothers and fathers who endorsed higher levels of couples satisfaction and dyadic coping would demonstrate higher rates of parental responsivity in the dyadic parent-child interactions (Pratt et al., 1992).

Method

Procedures and Measures

The data for the current study were collected as part of a larger study investigating multiple aspects of family relationships and parent and child behavior in families of children with FXS. Participants included 23 families of male children with FXS between the ages of 3;0 and 7;11 years, with a total of 69 participants including 23 fathers (22 biological fathers and one stepfather), 23 biological mothers, and 23 male children with FXS. See pages 5 – 6 for additional details on the participants in the current study.

Parents independently completed multiple questionnaires via REDCap (Harris et al., 2019; Harris et al., 2009), including the Symptom Checklist-90-Revised (SCL-90-R; Derogatis, 1994), the Parenting Stress Index – Fourth Edition, Short Form (PSI-4-SF; Abidin, 2012), the Couples Satisfaction Index (CSI-32; Funk & Rogge, 2007), and the Dyadic Coping Inventory (DCI; Bodenmann, 2008; Ledermann et al., 2010). The SCL-90-R measures mental health symptoms along multiple dimensions yielding a Positive Symptom Total, a Positive Symptom Distress Index, and a Global Severity Index (GSI). The GSI T-score was used in analyses. The PSI-4-SF measures parenting stress, providing subscale scores for Parental Distress, Parent-Child Dysfunctional Interaction, and Difficult Child. The Total Stress T-score was used in analyses. The CSI-32 measures couples satisfaction, with higher scores indicating higher levels of relationship satisfaction. Total raw scores were used in analyses. The DCI measures perceived communication and coping that occurs in relationships when one or both partners are experiencing stress. Higher scores indicate higher levels of dyadic coping. Total raw scores from this measure were used in analyses. See pages 7 – 10 for additional details regarding the measures used in the current study.

Mothers and fathers also separately engaged in a 12-minute dyadic play-based interaction with their child. The parent-child interactions served as the language samples for the current study. These interactions were recorded on different days of the study using secure video-based teleconferencing software (i.e., Skype for Business or Zoom). The interactions were transcribed using SALT (Systematic Analysis of Language Transcripts; Miller & Iglesias, 2008) and coded for the presence of responsive parental behaviors using a coding scheme adapted from Warren and colleagues (e.g., Warren et al., 2010). See page 11 – 13 for additional details regarding transcription and coding, including definitions and examples of the specific codes used in the current study.

Analysis Plan

All variables were visually inspected to check for model assumptions of normality and homoscedasticity of the residuals. 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 of the primary outcome variables for Aims 1 and 2 (i.e., parental responsivity and behavior management coded from mothers' and fathers' respective parent-child dyadic interactions) were reported. 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 parent utterances in the transcript. For example, a parent who had 144 responsive utterances, 17 behavior management utterances, and 227 total utterances would have a responsivity proportion of 144/227 = 0.63 and a behavior management proportion of 17/227 = 0.07.

Interspousal correlations were also reported to determine the degree of correspondence between the mothers' and fathers' responsive behaviors in the dyadic interactions. Then, intraclass correlation coefficients (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. Finally, a multilevel modeling (MLM) approach was used to manage the non-independence of the data collected from couples within families (Raudenbush & Bryk, 2002). In this approach, the data from each partner are nested within a dyad (Campbell & Kashy, 2002). Effect coding was used for parent sex (i.e., Male = 1 and Female = -1), and continuous predictors were grand-mean centered.

For Aim 1, separate models for parental responsivity and behavior management were specified. Given the substantial range in the data for parental responsivity and behavior management, and to better account for the quality of the parents' language as opposed to the quantity, proportion scores for the dependent variables were used instead of frequency scores. The strong and significant association between the variables for parenting stress and mental health challenges did not allow for them both to be included as predictors in the models for Aim 1; the parenting stress measure (PSI-4-SF Total T-score) was chosen because it was associated with parental responsivity for mothers, whereas the measure of mental health challenges was not associated with either parental responsivity or behavior management for mothers or fathers. Other predictors in the models for Aim 1 included parent sex and parent education.

As with Aim 1, separate models for parental responsivity and behavior management were specified for Aim 2, again using proportion scores for the dependent variables instead of frequency scores. The strong and significant association between the couples satisfaction or dyadic coping variables did not allow for them both to be included in the models for Aim 2; the couples satisfaction measure (CSI-32 Total raw score) was chosen as the primary predictor for Aim 2 as there was a marginally significant association between the CSI-32 and parental responsivity across all parents. Parent sex and parent education were also included as predictors in the models for Aim 2.

Results

Aim 1 was designed to examine relationships among maternal responsivity, paternal responsivity, and parent individual well-being (i.e., mental health challenges and parenting stress). Aim 2 was to examine relationships among maternal responsivity, paternal responsivity, and couple well-being (i.e., couples satisfaction and dyadic coping).

Table 1 displays descriptive statistics for the parental responsivity and behavioral management variables that were coded from the mother-child and father-child dyadic play-based interactions. For both parental responsivity and behavior management, frequency counts and proportions are reported. Paired samples t-tests and Wilcoxon signed-ranks tests (when appropriate) confirmed that there were no statistically significant differences between mothers' and fathers' use of responsive behaviors or behavior management strategies except for frequency of comments ($t(22) = 2.77$, $p = 0.011$) and the behavior management proportion score ($Z = -2.01$, $p = 0.044$). Mothers used comments more frequently than fathers, and fathers had a higher proportion score for behavior management compared to mothers.

Additionally, the differences between mothers' and fathers' frequency of redirects (Z = -1.69, p = 0.092) and zaps (Z = -1.78, p = 0.075) approached significance, with fathers using redirects and zaps more frequently than mothers.

Table 1

Mother-Father Comparisons of Parental Behavior During Dyadic Parent-Child Interactions

	Mean (*SD*) Range		ρ (p-value)
	Mothers	Fathers	Interspousal Correlation
Parental Responsivity			
Frequency of Comments	52.22 (21.69) 20 – 90	42.13 (22.44) 8 – 84	**0.71***** **(0.0001)**
Frequency of Requests for Verbal Compliance	39.17 (22.92) 5 – 96	38.52 (20.21) 3 – 72	**0.68***** **(0.0004)**
Frequency of Recodes	2.00 (2.00) 0 – 5	1.83 (2.55) 0 – 10	0.32 (0.135)
Total Parental Responsivity (Frequency)	93.39 (42.91) 26 – 176	82.47 (39.58) 11 – 149	**0.81***** **(0.00001)**
Total Parental Responsivity (Proportion of Total Utterances)	0.33 (0.14) 0.07 – 0.61	0.35 (0.15) 0.13 – 0.60	**0.69***** **(0.0003)**
Behavior Management			
Frequency of Requests for Behavioral Compliance	12.57 (12.25) 1 – 51	14.04 (11.59) 2 – 55	0.29 (0.177)
Frequency of Redirects	1.13 (1.42) 0 – 5	1.96 (1.87) 0 – 6	-0.05 (0.835)
Frequency of Zaps	0.74 (1.21) 0 – 5	1.57 (2.76) 0 – 13	0.03 (0.884)
Total Behavior Management (Frequency)	14.43 (12.41) 3 – 53	17.57 (12.78) 4 – 58	0.34 (0.116)
Total Behavior Management (Proportion of Total Utterances)	0.05 (0.04) 0.01 – 0.14	0.07 (0.04) 0.02 – 0.19	0.29 (0.177)

Note. * $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$.

Table 1 also displays interspousal correlations; given that some of the scores on these measures 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 mothers' and fathers' frequencies of comments and requests for verbal compliance, but not recodes. There were also significant correspondences between mothers' and fathers' responsivity frequency and proportion totals. No significant correspondences were found between mothers' and fathers' use of the individual behavior management strategies or the total frequency or proportion scores for behavior management.

Tables 2 and 3 displays correlations between measures of parent individual well-being (i.e., SCL-90-R Global Severity Index and PSI-4-SF Total Stress T-scores), couple well-being (i.e., CSI-32 and DCI total raw scores), and parent behavior (i.e., responsivity and behavior management) for mothers and fathers, respectively. Table 4 displays correlations between these parent variables combined across mothers and fathers. Table 2 shows that there was a significant correlation between parenting stress and responsivity for mothers as well as a marginally significant correlation between dyadic coping and parental responsivity; these relationships were not found for fathers. However, for parents overall, there was a significant correlation between parenting stress and responsivity (Table 4). Table 4 also shows that there was a marginally significant correlation between couples satisfaction and responsivity.

Table 2

Spearman Correlations for Maternal Variables

Variable	1.	2.	3.	4.	5.	6.
1. Responsivity	1.00					
2. Behavior Management	-0.16 (0.471)	1.00				
3. SCL-90-R[1]	-0.24 (0.277)	0.32 (0.138)	1.00			
4. PSI-4-SF[2]	**-0.45*** **(0.030)**	0.27 (0.207)	**0.77***** **(0.00001)**	1.00		
5. CSI-32[3]	0.12 (0.571)	-0.24 (0.261)	-0.32 (0.138)	**-0.49*** **(0.018)**	1.00	
6. DCI[4]	0.35$^{\sim}$ (0.097)	-0.20 (0.356)	-0.21 (0.325)	**-0.48*** **(0.021)**	**0.78***** **(0.00001)**	1.00

Note. Proportion scores used for Responsivity and Behavior Management variables. [1]SCL-90-R Global Severity Index T-score. [2]PSI-4-SF Total Stress T-score. [3]CSI-32 raw score. [4]DCI raw score.
$^{\sim}$ $p < 0.10$, * $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$.

Table 3

Spearman Correlations for Paternal Variables

Variable	1.	2.	3.	4.	5.	6.
1. Responsivity	1.00					
2. Behavior Management	-0.25 (0.250)	1.00				
3. SCL-90-R[1]	-0.08 (0.724)	0.25 (0.252)	1.00			
4. PSI-4-SF[2]	-0.33 (0.125)	0.23 (0.300)	**0.54**** **(0.008)**	1.00		
5. CSI-32[3]	0.33 (0.126)	-0.22 (0.315)	-0.17 (0.447)	**-0.68***** **(0.0004)**	1.00	
6. DCI[4]	-0.01 (0.968)	-0.04 (0.868)	-0.40$^{\sim}$ (0.061)	**-0.48*** **(0.021)**	**0.48*** **(0.019)**	1.00

Note. Proportion scores used for Responsivity and Behavior Management variables. [1]SCL-90-R Global Severity Index T-score. [2]PSI-4-SF Total Stress T-score. [3]CSI-32 raw score. [4]DCI raw score.
$^{\sim}$ $p < 0.10$, * $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$.

Table 4

Spearman Correlations for Combined Parental Variables

Variable	SCL-90-R[1]	PSI-4-SF[2]	CSI-32[3]	DCI[4]	Resp	BM
Responsivity	-0.15 (0.310)	**-0.41**** **(0.004)**	0.25$^{\sim}$ (0.090)	0.16 (0.291)	1.00	
Behavior Management	0.24 (0.111)	0.17 (0.271)	-0.20 (0.188)	-0.12 (0.443)	-0.17 (0.250)	1.00

Note. Proportion scores used for Responsivity and Behavior Management variables. [1]SCL-90-R Global Severity Index T-score. [2]PSI-4-SF Total Stress T-score. [3]CSI-32 raw score. [4]DCI raw score.
$^{\sim}$ $p < 0.10$, * $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$.

Aim 1 Models – Influence of Parent Well-Being on Parent 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 5 presents the results of the MLM analyses for Aim 1. The behavior management variable was log-transformed to reduce positive skew.

Table 5

Multilevel Model Results for Aim 1

	Parental Responsivity	Behavior Management
Fixed Effects		
Intercept	0.34^{***}	-3.04^{***}
	(0.02)	(0.12)
Parenting Stress	$-0.003^{\sim}$	$0.02^{\sim}$
	(0.002)	(0.01)
Parent Sex	0.01	0.24^{**}
	(0.20)	(0.08)
Parent Education	-0.01	0.09
	(0.01)	(0.07)
Random Effects		
Residual (σ^2_e)	0.01	0.29
Intercept (σ^2_{u0})	0.01	0.18
Goodness-of-fit		
AIC	-25.01	116.12
BIC	-14.04	127.09

Note. Standard errors in parentheses.
$^{\sim}$ $p < 0.10$, * $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$.

Prediction of Parental Responsivity

There was a marginally significant effect of parenting stress on parental responsivity ($p = 0.074$). Across all couples, when the other predictors were at their mean values, parents with greater than average parenting stress used a smaller proportion of responsive behaviors during the dyadic parent-child interactions. There were no significant main effects of parent sex or education on parental responsivity.

Prediction of Behavior Management

There was a marginally significant effect of parenting stress on behavior management ($p = 0.053$). Across all couples, when the other predictors were at their mean values, parents with greater than average parenting stress used a greater proportion of behavior management strategies during the dyadic parent-child interactions. There was also a significant main effect of parent sex on behavior management. In reference to the overall mean, when the other predictors were at their mean values, fathers used a greater proportion of behavior management strategies during the parent-child interactions than mothers. There was no significant main effect of parent education on behavior management.

Aim 2 Models – Influence of Couple Well-Being on Parent Behavior

Table 6 presents the results of the MLM analyses for Aim 2. As with Aim 1, the behavior management variable was log-transformed to reduce positive skew.

Table 6

Multilevel Model Results for Aim 2

	Parental Responsivity	Behavior Management
Fixed Effects		
Intercept	0.34***	-3.04***
	(0.02)	(0.12)
Couples Satisfaction	-0.0002	0.002
	(0.001)	(0.003)
Parent Sex	0.01	0.23**
	(0.01)	(0.09)
Parent Education	-0.01	0.05
	(0.01)	(0.08)
Random Effects		
Residual (σ^2_e)	0.02	0.34
Intercept (σ^2_{u0})	0.01	0.15
Goodness-of-fit		
AIC	-20.34	121.42
BIC	-9.37	132.40

Note. Standard errors in parentheses.
$^{\sim}$ $p < 0.10$, * $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$.

Prediction of Parental Responsivity

Unexpectedly, there were no significant main effects of couples satisfaction, parent sex, or parent education on parental responsivity.

Prediction of Behavior Management

Again, as with Aim 1, there was a significant main effect of parent sex on behavior management, suggesting that in reference to the overall mean, when the other predictors were at their mean value, fathers used a greater proportion of behavior management strategies during the parent-child interactions than mothers. However, there were no significant main effects of couples satisfaction or parent education on behavior management.

Discussion

The current study was designed to examine relationships between parental and couple characteristics and parent behavior during parent-child interactions. Findings suggest that mothers and fathers within families use similar levels of responsive strategies with their child. Specifically, comparisons of mothers' and fathers' behavior coded from the parent-child dyadic interactions demonstrated that there were significant correspondences between parents' use of responsive behaviors (i.e., comments, requests for verbal compliance, and recodes), with the exception of recodes. Total parental responsivity (measured both in frequency and in the proportion of the parents' total utterances) was also related for mothers and fathers. Mothers, however, used significantly more comments compared to fathers during the parent-child interactions. Maternal and paternal use of requests for verbal compliance, a parent question or statement that serves to elicit a spoken response from the child, were not significantly different, nor were there significant differences between mothers' and fathers' use of recodes, which are parent interpretations of the child's communication that extend the form of the child's act. Recodes were used very infrequently compared to comments and requests for verbal compliance. This finding may be due to the fact that when parents produced multiple utterances in succession, only the final utterance prior to either a three second pause or a child communication act was coded, consistent with past research on maternal responsivity in FXS (e.g., Brady et al., 2014; Warren et al., 2010; Wheeler et al., 2007). Therefore, the coding of parental behavior, as well as parents' use of recodes in particular, are likely to be affected by the child's language level.

No correspondences between mothers' and fathers' use of behavior management strategies (i.e., requests for verbal compliance, redirects, and zaps) were found in the current study. Additionally, there were marginally significant differences between parental use of both redirects and zaps, with fathers using these behaviors more frequently than mothers. Overall, fathers also used a higher proportion of behavior management strategies. However, for all parents, these behaviors were fairly low incidence, especially in comparison to responsive behaviors, which is consistent with past research on maternal behavior during mother-child interactions in families of children with FXS (e.g., Wheeler et al., 2007). Moreover, redirects and zaps occurred very infrequently (redirects: maternal M = 1.13 versus paternal M = 1.96; zaps: maternal M = 0.74 versus paternal M = 1.57). Requests for behavioral compliance, which were the most common behavioral management strategy used, tended to center around suggestions regarding play. For example, in one interaction, a father said to his child, "Let's see how tall we can build this tower." In another interaction, a mother said to her child, "Pour it into my dish here." These types of parent utterances encouraged meaningful and interactive play and engagement in the interaction. Future studies focused on other developmental outcomes in these families should investigate these parent behaviors further, as well as differences between maternal and paternal use of behavior management strategies.

There were some interesting differences in the correlations between parental responsivity and mothers' versus fathers' measures of individual and couple functioning. Specifically, parental responsivity was related to parenting stress for mothers, but not for fathers. This finding may be related to the fact that more mothers compared to fathers reported clinically significant levels of stress on the PSI-4-SF. There was also a marginally significant relationship between dyadic coping and parental responsivity for mothers but not fathers. More research is needed to understand potential differences between mothers and fathers in the relationships between individual well-being and couple well-being, both in families of children with neurodevelopmental disabilities as well in families of neurotypical children.

Moreover, when the data for mothers and fathers were combined, there was a significant relationship between parenting stress and parental responsivity and a marginally significant relationship between couples satisfaction and parental responsivity. Interestingly, no significant relationships were found between behavior management and the measures of parent and couple functioning, which suggests that parental directive behaviors may be influenced by other factors or not related to individual or couple well-being, possibly including factors related to the child's behavior or developmental level.

In the multilevel models for Aim 1, parenting stress was found to associate with both parental responsivity and behavior management; however, the main effects were only marginally significant in each of these models. Wheeler et al. (2007) found that maternal stress was a significant predictor of the total number of maternal behaviors (i.e., maintaining and directing behaviors) exhibited during mother-child interactions, demonstrating that mothers with higher levels of stress engaged in fewer interactions with their child. Sterling et al. (2013), however, did not find maternal stress to be a significant predictor of maternal responsivity when child developmental level, maternal depressive symptoms, and maternal IQ were also included in the model as predictors. Wheeler et al. (2007) used the Total Stress score from the PSI to predict maternal responsivity (as was done in the current study), whereas Sterling et al. (2013) used the Parental Distress subscale score of the PSI. The difference in findings between Wheeler et al. (2007) and the current study may be related to the fact that 52% of mothers in the study by Wheeler et al. (2007) reported experiencing clinically significant levels of parenting stress compared to approximately only 20% of parents in the current study.

Additionally, in contrast to Sterling et al. (2013), parental education did not significantly predict either parental responsivity or behavior management in the present study. However, this finding was likely due to the limited variability in education status in the current sample, given that the majority of parents in the study had either a bachelor's degree, master's degree, or other advanced degree. Future studies with more diverse samples could investigate the contributions of parental education on parent behavior in these families, or measure parent IQ and examine IQ as a predictor of parent behavior.

Parent sex was not found to associate with parental responsivity in the multilevel model for Aim 1; however, there was a significant main effect of parent sex on behavior management in the analyses for both Aims 1 and 2. In particular, fathers used a higher proportion of behavior management strategies compared to mothers during the parent-child interactions. Couples satisfaction did not predict either parental responsivity or behavior management in the models for Aim 2 and couples satisfaction was only marginally significantly correlated with parental responsivity (Table 4). However, couples satisfaction in this sample was negatively skewed, with a majority of parents reporting moderate to high levels of satisfaction; thus, a lack of variability in couples satisfaction could explain the lack of a correlation with responsivity. Parents who were satisfied with their relationships may have been more likely to participate in the current study compared to dissatisfied couples. Therefore, future studies should examine these relationships further in larger samples given past studies that demonstrate the influence of parent and couple functioning on parent behavior in both dyadic and triadic (i.e., mother-father-child) interactions (e.g., Kouros et al., 2014; Pratt et al., 1992; Stroud et al., 2011).

Limitations and Future Directions

One significant limitation of the current study is the small sample size, which may have influenced the power to detect significant main effects of both parenting stress and couples satisfaction on parenting behavior. Moreover, the sample lacked diversity in terms of race/ethnicity—with the majority of the participants identifying as White and not Hispanic or Latinx—and parent education, with approximately 70% of both mothers and fathers having a bachelor's degree, master's degree, or other advanced degree.

Another limitation is the use of only one sampling context for the parent-child interactions. Past studies have gathered and analyzed data from multiple parent-child interaction contexts, including free play, unstructured naturalistic activities (e.g., folding laundry, putting dishes away), book reading, and making and eating a snack together (e.g., Brady et al., 2014; Warren et al. 2010). In these studies, parental responsivity was coded from a total of approximately 25 minutes of parent-child interactions across these contexts. The use of additional contexts or longer interactions could have led to more variability in the data within and between families, which may have increased the ability to detect significant relationships between individual and couple functioning and parent behavior in the current study.

The use of frequencies versus proportions to analyze the effects of parental input on child outcomes in past research is varied (Rowe & Snow, 2020), with several studies reporting is frequencies (e.g., Landry et al, 2001; McDuffie & Yoder, 2010) and several other studies reporting proportions (e.g., Ambrose et al., 2015; Bornstein et al., 1992; Lorang et al., 2020). Importantly, mothers in the current study provided more linguistic input compared to fathers (i.e., had a greater total number of utterances overall; maternal M = 288.52, SD = 62.22 versus paternal M = 240.30, SD = 82.97), which is consistent with past research on parental input in families of neurotypical children (Davidson & Snow, 1996; Pancsofar & Vernon-Feagans, 2006; Shapiro et al., 2021). Mothers also used significantly more comments than fathers (maternal M = 52.22, SD = 21.69 versus paternal M = 42.13, SD = 22.44), but the difference in the proportion of comments used by mothers compared to fathers was not significantly different (maternal *M* = 0.185, *SD* = 0.07 versus paternal *M* = 0.178, *SD* = 0.08), nor was the difference in the overall proportion of responsive behaviors used (maternal *M* = 0.33, *SD* = 0.14 versus paternal *M* = 0.35, *SD* = 0.15). The current study sought to examine differences in the quality of maternal and paternal input as opposed to the quantity, and therefore used proportions instead of frequencies. However, future studies could examine multiple metrics of parental responsivity (e.g., frequency, proportion of total utterances, rate per minute) to better understand the effects of potential differences in maternal versus paternal input on various outcomes.

Moreover, although a great deal of past research has demonstrated that children develop language through interactions with their parents, the majority of this work conducted in families of children with developmental disabilities has neglected the fact that these interactions are dynamic and often involve more than one communicative partner at a time. That is, children spend a substantial amount of time engaging in dyadic interactions with their mothers and fathers as well as in triadic mother-father-child interactions (McHale & Fivaz-Depeursinge, 1999). The triad is comprised of multiple family subsystems (i.e., the mother-child dyad, father-child dyad, and mother-father dyad) and thereby is a more complex and diverse environment than any of the individual dyadic partnerships (Lindsey & Caldera, 2006; Stoneman & Brody, 1981). Therefore, determining the variability in child and parent behavior that arises as a function of whether the interaction is dyadic or triadic, as well as the individual and dyadic-level variables that contribute to that variability, are other areas for future research in families affected by FXS.

Additionally, the coding procedures used in the current study were such that certain responsive parental behaviors may have been missed if the parents' utterance was not followed by a child utterance or a three second pause. For example, parent recodes were very low frequency. However, parents may have been recoding the child's communication more frequently than the data in the current study reflect, but immediately saying something else following the recode, thereby interfering with that behavior being coded. Indeed, this could be true for all types of parent behavior coded during these interactions. Future studies should investigate the various types of parentally responsive behavior further with different coding methods, as well as how the use of different types of behavior may change over time as the child develops.

Another important consideration regarding the findings of the current study is that nearly all of these data were collected during the COVID-19 pandemic. In addition to widespread concerns about the coronavirus, parents may have been experiencing elevated levels of stress during this time due to loss of employment, social isolation, additional caregiving responsibilities, and the increased burden of managing the child's educational and therapeutic programs (Chan & Fung, 2021; Neece et al., 2020). Despite the relatively high levels of parental stress reported in the current study, the majority of parents in the sample reported average to above average levels of couples satisfaction. In fact, only approximately 20% of parents reported notable relationship dissatisfaction, which likely limited our ability to detect a significant effect of couples satisfaction on parental responsivity. Future studies should continue to examine relationships between parent and couple functioning and parent behavior in larger and more diverse samples.

Conclusions

The findings from the current study provide preliminary evidence that parenting stress may influence the responsive behavior of both mothers and fathers during parent-child interactions in families of children with FXS. Although only a marginally significant main effect of parenting stress on parental responsivity was detected, these findings still suggest that families would likely benefit from interventions aimed at reducing parent stress (e.g., Neece, 2014; Neece et al., 2019), especially given the prevalence of elevated parenting stress in parents of children with FXS (e.g., Johnston et al., 2003; McCarthy et al., 2006). Future studies should continue to investigate individual and couple functioning for both mothers and fathers of children with FXS as well as the influence of these characteristics on parent behavior.

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Study 3: Parental Responsivity and Child Communication During Mother-Child and Father-Child Interactions in Fragile X Syndrome

Abstract

Past research shows that parentally responsive behavior positively influences language development in both neurotypical children and children with intellectual and developmental disabilities, including those with fragile X syndrome (FXS), but the majority of these studies have focused exclusively on the mother-child relationship. Therefore, very little is known about how paternal behavior compares to maternal behavior or how paternal behavior relates to child outcomes. 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. Participants were 23 families of young boys with FXS between the ages of 3 and 7 years. Results indicated that mothers and fathers used similar rates of responsive behaviors during parent-child interactions, and that parental responsivity was positively associated with child language performance, including talkativeness and lexical diversity. Parental behavior, however, was not associated with child syntactic complexity. Moreover, findings also indicated that older children and children with higher levels of adaptive behavior had parents who used higher rates of responsive behaviors. Additionally, fathers used higher rates of behavior management strategies compared to mothers, and this type of parental behavior was not associated with child language performance. Overall, this study provides evidence that interventions focused on increasing parental responsiveness would be beneficial for these families and that these interventions should be delivered early given the association between responsivity and child age. The similarities in parental behavior across mother-child and father-child interactions suggests that including both parents in the intervention would likely lead to better outcomes for families.

Keywords: fragile X syndrome, parental responsivity, child language, behavior management

Individuals with fragile X syndrome (FXS) experience significant delays in multiple domains of language (Abbeduto et al., 2007). Language is one of the most important developmental domains to understand and target for intervention in these individuals 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). In recent years, research on maternal responsivity in FXS suggests that both early and sustained responsivity are critically important for child outcomes across a variety of domains (e.g., Brady et al., 2020; Brady et al., 2014; Warren et al., 2017; Warren et al., 2010). Moreover, recent parent-implemented language interventions for children with FXS between the ages of two and 17 years have shown that parents are able to learn and implement targeted responsive strategies 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, the majority of past research on the role of responsive parenting in FXS, and on parent-implemented language interventions, has focused exclusively on the mother-child dyad. The goals of the current study are (1) to examine relationships between parental responsivity and child language performance in young boys with FXS; (2) to examine relationships between characteristics of the child and both parental responsivity and child language performance; and (3) to expand the focus beyond the mother-child dyad to include examinations of these relationships in father-child dyads.

Behavioral Phenotype of *FMR1*-Associated Conditions

Fragile X syndrome (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 1 in 7,143 males are affected by the full mutation compared to only approximately 1 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).

Other phenotypic characteristics of FXS, all of which are likely to interfere with language learning and social interaction, include hyperactivity (Baumgardner et al., 1995), deficits 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 symptoms of ASD, with as many as 50-60% of males receiving a diagnosis of ASD when diagnostic tools designed to measure symptoms of ASD in the general population are used (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., 2015).

Only females can transmit the *FMR1* full mutation to the next generation. 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 (FXPOI) and fragile X-associated tremor ataxia syndrome (FXTAS), a late-onset neurogenerative disease that affects both male and female premutation carriers (Visootsak et al., 2014). Females with the *FMR1* premutation are 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 deficits in executive functioning, attention, working memory, arithmetic, and various 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 a disability. These challenges include high levels of child challenging behaviors, low levels of adaptive behaviors, an increased financial burden, and social isolation (Minnes et al.,Weiss, 2015; Tint & Weiss, 2015).

The cumulative effects of the factors affecting female carriers may constrain the development of a warm and responsive mother-child relationship with subsequent negative impacts on optimal 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 relationship and the father-child relationship and thereby the child's development across multiple domains.

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 considered to be 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 the child becomes more communicative and socially engaged, responsive parents modify their behavior and adapt to their child's developing abilities and interests by maintaining their child's focus of attention and following the lead of the child through behaviors such as commenting and recasting on the child's current 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 six and twelve months after participation in two different prelinguistic communication interventions (Yoder & Warren, 2001). Moreover, McDuffie & Yoder (2010) found that certain types of parental verbal responsiveness predicted early vocabulary acquisition in young children with ASD; specifically, parents' use of follow-in comments, follow-in directives, and expansions of child communication acts. 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 the child's linguistic development, particularly for those children in the sample who were older and more communicative. That is, the extent to which mothers are able to successfully adapt to their children's linguistic growth by increasing their use of facilitative behaviors that match their child's current levels 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 three 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 ASD symptoms and nonverbal developmental level. Similarly, in another study of these mother-child dyads, Brady et al. (2014) found that sustained responsivity measured across four years predicted later receptive and expressive vocabulary development up to nine years of age. In a 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 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 that maternal responsivity in FXS is not only important in early childhood (Warren et al., 2010) and middle childhood (Brady et al., 2014), but that sustaining responsivity has a positive influence on language development even throughout adolescence and, therefore, 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 was found to have a significant and positive impact on growth in child Communication scores, even after controlling for ASD symptoms and developmental level. In addition, maternal responsivity predicted trajectories of Socialization and Daily Living Skills, but to a lesser extent than Communication skills. 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 in 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 current study was designed to replicate some of these past findings in a new sample of mother-child dyads, and to extend the investigation to also include father-child dyads.

Theoretical Framework

Both the transactional model (Sameroff & Chandler, 1975) and family systems theory (Cox & Paley, 1997) 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 "any individual family member is inextricably embedded in the larger family system and can never be fully understood independent of the context of that system" (Cox & Paley, 1997, p. 246). 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. According to Laursen and Collins (2009), "bidirectional models imply that parent behaviors are both the cause and the consequence of child behaviors" (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 such that the parent and child are an interdependent unit (e.g., Belsky, 1984; Neece et al., 2012; Sameroff & Mackenzie, 2003; Sameroff, 2009). 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). The current study examined relationships between parent behavior, child behavior, and child characteristics in both mother-child and father-child interactions.

Current study

The current study was designed to examine relationships between parental responsivity and child language, as well as the ways in which child characteristics affect parent behavior and relate to language performance, in both mother-child and father-child dyads. The first aim 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 symptoms, and adaptive behavior) and child language. We hypothesized that higher rates of parental 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 parental responsivity. We hypothesized that higher levels of child adaptive behavior as well as fewer challenging behaviors and ASD symptoms would be associated with higher rates of parental responsivity (Warren et al., 2010; Brady et al., 2014).

Method

Procedures 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. Participants included 23 families of male children with FXS between the ages of 3;0 and 7;11 years, with a total of 69 participants including 23 fathers (22 biological fathers and one stepfather), 23 biological mothers, and 23 male children with FXS. See pages 5 – 6 for additional details on the participants in the current study.

In order to address the aims of this study, mothers and fathers independently completed two questionnaires via REDCap (Harris et al., 2019; Harris et al., 2009) to assess child characteristics, including the Aberrant Behavior Checklist-Community, 2nd edition (ABC-2; Aman & Singh, 2017) and the Social Responsiveness Scale, 2nd edition (SRS-2; Constantino & Gruber, 2012). One parent also completed the Vineland Adaptive Behavior Scales, 3rd edition (Vineland-3; Sparrow, Cicchetti, & Saulnier, 2016) as an interview to assess the child's adaptive behavior.

The ABC-2 measures challenging behaviors of individuals with developmental disabilities in several domains. Total raw scores from the FXS-specific subscale scoring (Sansone et al., 2012) were used in analyses. The SRS-2 measures social impairments commonly associated with ASD, providing DSM-5 compatible subscale scores for Social Communication and Interaction (SCI) and Restricted

Interests and Repetitive Behavior (RRB), as well as a Total T-score. Total T-scores from the SRS-2 were used in analyses. 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.

Mothers and fathers also separately engaged in 12-minute play-based interactions with their child. The parent-child interactions served as the language samples for the current study. These interactions were transcribed using SALT (Systematic Analysis of Language Transcripts; Miller & Iglesias, 2008) and coded for parental behavior using a coding scheme adapted from Warren and colleagues (e.g., Warren et al., 2010; see page 13 for definitions and examples of the specific codes used in the current study). Measures of parent and child language were also obtained from the SALT transcripts, including total number of utterances (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). See pages 9 – 13 for additional details regarding the measures and procedures used in the current study.

Analysis Plan

All variables were visually inspected to check for model assumptions of normality and homoscedasticity of the residuals. 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 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. 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 (1) maternal ratings of the child and mother-child interaction variables, (2) paternal ratings of the child and father-child interaction variables, and (3) 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 included the following child language measures obtained from the SALT transcripts of the dyadic play-based language samples: total number of 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 non-independence of the data collected from different parent-child dyads within families. In this approach, the data from each dyad is 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, intraclass correlation coefficients (ICCs) were calculated to estimate the proportion of the total variation in the child language measures (i.e., total number of utterances, NDW, and MLUm) that exists between versus within families. Then, separate models for child total number of 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 associations between the variables for child ASD symptoms, child challenging behavior, and child adaptive behavior; however, there was not a significant association 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 that were used in Aim 1 were used in Aim 2 to predict parent behavior. Parent sex was also included as a predictor to examine whether there were differences in parent behavior between the mother-child and father-child interactions.

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 relationships between child characteristics and child language performance. Aim 2 was designed to examine relationships between child characteristics and parental responsivity. Table 1 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, mothers' and fathers' language, or mothers' and fathers' use of responsivity or behavior management strategies except for parent talkativeness (i.e., total number of utterances; $t(22) = 3.22$, $p = 0.004$), parent lexical diversity (i.e., NDW; $t(22) = 4.29$, $p = 0.0003$), and the behavior management proportion score ($Z = -2.01$, $p = 0.031$). Mothers had a greater number of total utterances and different words compared to fathers, and fathers had a higher proportion score for behavior management strategies compared to mothers.

Table 1 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 found between mothers' and fathers' use of the individual behavior management strategies or the total frequency or proportion scores for behavior management.

Table 1

Comparisons of Parental Behavior and Child Language During Dyadic Parent-Child Interactions

	Mean (*SD*) Range		ρ (*p*-value)
	Mother-Child Interaction	Father-Child Interaction	Interspousal Correlation
Parent Language			
Total Utterances	288.52 (62.22) 150 – 405	240.30 (82.97) 85 – 450	**0.49*** **(0.017)**
Number of Different Words	211.48 (46.71) 126 – 307	171.57 (49.46) 74 – 271	**0.55**** **(0.007)**
Mean Length of Utterance - Morphemes	3.36 (0.51) 2.21 – 4.53	3.41 (0.50) 2.43 – 4.25	**0.42*** **(0.047)**
Total Parental Responsivity (Frequency)	93.39 (42.91) 26 – 176	82.48 (39.58) 11 – 149	**0.81***** **(0.00001)**
Total Parental Responsivity (Proportion of Total Utterances)	0.33 (0.14) 0.07 – 0.61	0.35 (0.15) 0.13 – 0.60	**0.69***** **(0.0003)**
Total Behavior Management (Frequency)	14.43 (12.41) 5 – 53	17.57 (12.78) 4 – 58	0.34 (0.116)
Total Behavior Management (Proportion of Total Utterances)	0.05 (0.04) 0.01 – 0.14	0.07 (0.04) 0.02 – 0.19	0.29 (0.177)
Child Language			
Total Utterances	132.17 (87.29) 6 – 307	138.13 (89.34) 1 – 303	**0.81***** **(0.00001)**
Number of Different Words	68.13 (48.04) 1 – 154	71.09 (55.61) 1 – 178	**0.88***** **(0.00001)**
Mean Length of Utterance - Morphemes	1.70 (0.55) 1.00 – 2.75	1.82 (0.77) 1.00 – 3.88	**0.90***** **(0.00001)**

Note. $^{\sim}p < 0.10$, $^{*}p < 0.05$, $^{**}p < 0.01$, $^{***}p < 0.001$.

Tables 2 and 3 displays correlations between measures of parent behavior, child language, and child characteristics for mothers and fathers, respectively. Table 4 displays correlations between these variables combined across parents. Table 2 shows that there were significant positive correlations between maternal responsivity and the following variables: child age, child total utterances, child NDW, child MLUm, and the Vineland-3 Daily Living Skills and Socialization domains as well as the Vineland-3 Adaptive Behavior Composite score. There were no significant correlations between maternal behavior management and child language or child characteristics with the exception of a significant correlation between maternal behavior management and the Vineland-3 Socialization domain score.

Table 2

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 (0.471)	1.00						
3. Child Age	**0.42* (0.0495)**	0.06 (0.799)	1.00					
4. Child Total Utterances	**0.81*** (0.00001)**	0.18 (0.399)	**0.47* (0.025)**	1.00				
5. Child NDW[1]	**0.84*** (0.00001)**	-0.01 (0.993)	**0.47* (0.023)**	**0.94*** (0.00001)**	1.00			
6. Child MLUm[2]	**0.68*** (0.0004)**	-0.10 (0.666)	**0.47* (0.025)**	**0.75*** (0.00001)**	**0.89*** (0.00001)**	1.00		
7. Challenging Behavior[3]	-0.03$^{\sim}$ (0.089)	0.37$^{\sim}$ (0.080)	0.09 (0.687)	0.23 (0.290)	0.16 (0.468)	0.19 (0.393)	1.00	
8. ASD Symptoms[4]	-0.27 (0.209)	0.40$^{\sim}$ (0.059)	0.36$^{\sim}$ (0.093)	-0.05 (0.805)	-0.20 (0.369)	-0.17 (0.450)	**0.47* (0.023)**	1.00
9. Commun-ication[5]	0.41$^{\sim}$ (0.054)	-0.21 (0.343)	-0.25 (0.257)	0.39$^{\sim}$ (0.070)	**0.44* (0.036)**	**0.45* (0.031)**	-0.10 (0.653)	**-0.63 (0.001)**
10. Daily Living Skills[6]	**0.43* (0.043)**	-0.27 (0.209)	-0.17 (0.438)	**0.45* (0.031)**	**0.52* (0.011)**	0.40$^{\sim}$ (0.059)	-0.24 (0.273)	**-0.65 (0.0007)**
11. Social-ization[7]	**0.48* (0.021)**	**-0.43* (0.042)**	-0.19 (0.378)	0.34 (0.118)	**0.43* (0.041)**	0.39$^{\sim}$ (0.066)	-0.36$^{\sim}$ (0.094)	**-0.83 (0.00001)**
12. Vineland ABC[8]	**0.46* (0.026)**	-0.21 (0.326)	-0.25 (0.259)	**0.45* (0.032)**	**0.50* (0.015)**	**0.43* (0.039)**	-0.22 (0.315)	**-0.78 (0.00001)**

Note. Proportion scores used for Responsivity and Behavior Management variables. [1]NDW = number of different words. [2]MLUm = mean length of utterance in morphemes. [3]ABC-2 Total raw score. [4]SRS-2 Total T-Score. [5]Vineland-3 Communication domain standard score. [6]Vineland-3 Daily Living Skills domain standard score. [7]Vineland-3 Socialization domain standard score. [8]Vineland-3 Adaptive Behavior Composite score.

$^{\sim}$ $p < 0.10$, * $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$.

Table 3

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 (0.250)	1.00						
3. Child Age	0.27 (0.213)	0.29 (0.173)	1.00					
4. Child Total Utterances	**0.83***** **(0.00001)**	0.05 (0.809)	0.33 (0.130)	1.00				
5. Child NDW[1]	**0.81***** **(0.00001)**	0.04 (0.849)	0.40$^{\sim}$ (0.061)	**0.94***** **(0.00001)**	1.00			
6. Child MLUm[2]	**0.76***** **(0.00001)**	-0.07 (0.757)	**0.46*** **(0.025)**	**0.85***** **(0.00001)**	**0.93***** **(0.00001)**	1.00		
7. Challenging Behavior[3]	-0.32 (0.136)	-0.20 (0.356)	-0.28 (0.198)	**-0.47*** **(0.024)**	**-0.46*** **(0.026)**	**-0.43*** **(0.040)**	1.00	
8. ASD Symptoms[4]	**-0.47*** **(0.023)**	0.11 (0.632)	-0.10 (0.635)	**-0.47*** **(0.023)**	**-0.42*** **(0.046)**	**-0.45*** **(0.032)**	**0.66***** **(0.0007)**	1.00
9. Commun-ication[5]	**0.62**** **(0.002)**	-0.26 (0.236)	-0.25 (0.257)	**0.52*** **(0.011)**	**0.57**** **(0.005)**	**0.54**** **(0.007)**	-0.07 (0.748)	-0.27 (0.211)
10. Daily Living Skills[6]	**0.61**** **(0.002)**	-0.14 (0.536)	-0.17 (0.438)	**0.55**** **(0.007)**	**0.59**** **(0.003)**	**0.47*** **(0.025)**	-0.21 (0.342)	-0.23 (0.290)
11. Social-ization[7]	**0.60**** **(0.003)**	-0.343 (0.109)	-0.19 (0.378)	**0.48*** **(0.021)**	**0.49*** **(0.019)**	**0.48*** **(0.022)**	-0.28 (0.203)	**-0.43*** **(0.038)**
12. Vineland ABC[8]	**0.68**** **(0.001)**	-0.28 (0.200)	-0.25 (0.259)	**0.59**** **(0.003)**	**0.61**** **(0.002)**	**0.53**** **(0.009)**	-0.227 (0.297)	-0.40$^{\sim}$ (0.058)

Note. Proportion scores used for Responsivity and Behavior Management variables. [1]NDW = number of different words. [2]MLUm = mean length of utterance in morphemes. [3]ABC-2 Total raw score. [4]SRS-2 Total T-Score. [5]Vineland-3 Communication domain standard score. [6]Vineland-3 Daily Living Skills domain standard score. [7]Vineland-3 Socialization domain standard score. [8]Vineland-3 Adaptive Behavior Composite score.
$^{\sim}p < 0.10$, $^{*}p < 0.05$, $^{**}p < 0.01$, $^{***}p < 0.001$.

Table 3 shows that there were significant correlations between paternal responsivity and the following variables: child total utterances, child NDW, child MLUm, and child ASD symptoms, as well as all Vineland-3 domain (i.e., Communication, Daily Living Skills, and Socialization) 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 4

***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 (0.250)	1.00						
3. Child Age	**0.34*** **(0.019)**	0.17 (0.271)	1.00					
4. Child Total Utterances	**0.82***** **(0.00001)**	0.14 (0.356)	**0.40**** **(0.006)**	1.00				
5. Child NDW[1]	**0.83***** **(0.00001)**	0.03 (0.821)	**0.43**** **(0.002)**	**0.93***** **(0.00001)**	1.00			
6. Child MLUm[2]	**0.73***** **(0.00001)**	-0.06 (0.703)	**0.48***** **(0.0007)**	**0.79***** **(0.00001)**	**0.91***** **(0.00001)**	1.00		
7. Challenging Behavior[3]	-0.19 (0.218)	0.04 (0.797)	-0.08 (0.610)	-0.14 (0.368)	-0.15 (0.314)	-0.13 (0.404)	1.00	
8. ASD Symptoms[4]	**-0.39**** **(0.007)**	0.16 (0.277)	0.12 (0.419)	-0.28 (0.063)	**-0.30*** **(0.046)**	**-0.30*** **(0.044)**	**0.58***** **(0.00001)**	1.00
9. Communication[5]	**0.50***** **(0.0004)**	-0.21 (0.170)	-0.25 0.100	**0.44**** **(0.002)**	**0.51***** **(0.0003)**	**0.49***** **(0.0006)**	-0.10 (0.499)	**-0.45**** **(0.002)**
10. Daily Living Skills[6]	**0.51***** **(0.0003)**	-0.18 (0.239)	-0.17 (0.258)	**0.49***** **(0.0005)**	**0.57***** **(0.00001)**	**0.43**** **(0.003)**	-0.229 (0.126)	**-0.44**** **(0.002)**
11. Socialization[7]	**0.53***** **(0.0002)**	**-0.35*** **(0.016)**	-0.19 (0.199)	**0.41**** **(0.004)**	**0.46**** **(0.001)**	**0.42**** **(0.004)**	**-0.33*** **(0.027)**	**-0.64***** **(0.00001)**
12. Vineland ABC[8]	**0.56***** **(0.00001)**	-0.21 (0.167)	-0.25 (0.100)	**0.52***** **(0.0002)**	**0.56***** **(0.00001)**	**0.47**** **(0.0009)**	-0.24 (0.115)	**-0.59***** **(0.00001)**

Note. Proportion scores used for Responsivity and Behavior Management variables. [1]NDW = number of different words. [2]MLUm = mean length of utterance in morphemes. [3]ABC-2 Total raw score. [4]SRS-2 Total T-Score. [5]Vineland-3 Communication domain standard score. [6]Vineland-3 Daily Living Skills domain standard score. [7]Vineland-3 Socialization domain standard score. [8]Vineland-3 Adaptive Behavior Composite score.
$^{\sim} p < 0.10$, $^{*} p < 0.05$, $^{**} p < 0.01$, $^{***} p < 0.001$.

In the mother-child interactions, there were significant correlations between child age and the three measures of child language. However, in the father-child interactions, child age was associated with child MLUm, but not child total utterances or NDW. Additionally, in the father-child interactions, the child language measures were associated with paternal ratings of the child's challenging behavior and ASD symptoms as well as the three Vineland-3 domain scores and the Adaptive Behavior Composite score. In the mother-child interactions, the child language measures were not associated with maternal ratings of the child's challenging behavior or ASD symptoms. However, child talkativeness was associated with the Vineland-3 Daily Living Skills domain and Adaptive Behavior Composite score, child lexical diversity was associated with all Vineland-3 domain scores and the Adaptive Behavior Composite score, and child syntactic complexity was associated with the Vineland-3 Communication domain score and the Adaptive Behavior Composite score.

Table 4 shows that when the maternal and paternal variables are analyzed together, parental responsivity was associated with child age, ASD symptoms, 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 symptoms, or adaptive behavior. All child language measures were associated with adaptive behavior, and child lexical diversity and syntactic complexity were associated with ASD symptoms.

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 5 presents the results of the MLM analyses for Aim 1. The lexical diversity and syntactic complexity variables were square-root-transformed. Both of these transformations were done to reduce positive skew.

Table 5

Multilevel Model Results for Aim 1

	Child Talkativeness (Total Utterances)	Child Lexical Diversity (NDW)	Child Syntactic Complexity (MLUm)
Fixed Effects			
Intercept	135.15***	7.49***	1.30***
	(9.79)	(0.40)	(0.03)
Parental Responsivity	276.81***	8.21***	0.31
	(57.89)	(1.98)	(0.22)
Challenging Behavior	0.19	-0.0004	-0.0001
	(0.24)	(0.01)	(0.001)
Adaptive Behavior	2.51*	0.16***	0.01***
	(0.98)	(0.04)	(0.003)
Child Age	20.60**	1.07**	0.09***
	(7.67)	(0.31)	(0.02)
Parent Sex	0.47	-0.06	0.01
	(3.82)	(0.13)	(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. Standard errors in parentheses.
$^{\sim} p < 0.10$, $^{*} p < 0.05$, $^{**} p < 0.01$, $^{***} p < 0.001$.

Prediction of Child Talkativeness – Total Number of Utterances

As expected, there was a significant main effect of parental responsivity on child talkativeness. Across all dyads, when the other predictors were at their mean values, 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. Across all dyads, when the other predictors were at their mean values, 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, suggesting that across all dyads, when the other predictors were at their mean values, 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 – Number of Different Words

As with child talkativeness, there were significant main effects of parental responsivity, child adaptive behavior, and child age on child lexical diversity, but no significant main effects of either challenging behavior or parent sex on child lexical diversity. As such, across all dyads, when the other predictors were at their mean values, 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 at their mean values, 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 at their mean values, older children had higher levels of lexical diversity than younger children.

Prediction of Child Syntactic Complexity – Mean Length of Utterance in Morphemes

Unlike the other child language measures, there was not a significant main effect of parental responsivity on syntactic complexity. There were, however, significant main effects of both child adaptive behavior and child age on syntactic complexity. Across all dyads, when the other predictors were at their mean values, children with above average levels of adaptive behavior had higher levels of syntactic complexity compared to children with below average levels of adaptive behavior. Additionally, across all dyads, when the other predictors were at their mean values, 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, lexical diversity, or syntactic complexity. 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 6 presents the results of the MLM analyses for Aim 2. The behavior management variable was log-transformed to reduce positive skew.

Table 6

Multilevel Model Results for Aim 2

	Parental Responsivity	Behavior Management
Fixed Effects		
Intercept	0.34^{***}	-3.04^{***}
	(0.02)	(0.10)
Challenging Behavior	-0.0001	0.001
	(0.0006)	(0.004)
Adaptive Behavior[1]	0.008^{***}	-0.02^{*}
	(0.002)	(0.01)
Child Age	0.05^{***}	0.02
	(0.01)	(0.08)
Parent Sex	0.01	0.25^{**}
	(0.01)	(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. Standard errors in parentheses. [1]The Vineland-3 Socialization domain standard score was used to predict behavior management instead of the Vineland-3 Adaptive Behavior Composite score.
$^{\sim} p < 0.10$, $^{*} p < 0.05$, $^{**} p < 0.01$, $^{***} p < 0.001$.

Prediction of Parental Responsivity

As expected, there was a significant main effect of child adaptive behavior on parental responsivity. Across all dyads, when the other predictors were at their mean values, 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. Across all dyads, when the other predictors were at their mean values, 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.

Prediction of Behavior Management

There was a significant main effect of child adaptive behavior (i.e., socialization) on behavior management. Across all dyads, when the other predictors were at their mean values, 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. In reference to the overall mean, when the other predictors were at their mean values, 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, with no discernable differences between child language performance in the mother-child versus the 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 correspondences between mothers' and fathers' language use, mothers were more talkative (i.e., had a higher total number of utterances) and demonstrated greater lexical diversity (i.e., had a higher total number of different words) 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 the frequency of maternal and paternal input during dyadic parent-child interactions have been reported in families of children with FXS. Importantly, these differences in overall maternal and paternal input did not lead to any significant differences in child language performance. However, because only 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 or 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 mother compared to fathers. For example, child ASD symptoms (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 symptoms 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 symptoms 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 the other two language measures (i.e., talkativeness and 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 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 parent-implemented language interventions (e.g., McDuffie et al. 2018). For example, Komesidou et al. (2017), who examined the longitudinal trajectory of expressive syntax over three years in children with FXS, found significant syntactic growth over time, but maternal responsivity did not predict syntactic outcomes. The 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.

Moreover, in the current study, parent behavior management did not predict any of the child language measures, demonstrating the importance of certain kinds 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., 2020, Brady et al., 2014; Warren et al., 2017; Warren et al., 2010). Importantly, the findings of the current study demonstrate that paternal responsivity is important for child language performance as well, especially given that no differences were found in the child language measures between the mother-child and father-child interactions. 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 impact 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 no significant main effect of child challenging behavior on either parental responsivity or behavior management. However, in the current study, children 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 that found that the child's developmental level (as measured by the Mullen Scales of Early Learning; Mullen 1995) strongly influenced maternal responsivity (Sterling et al., 2013). Moreover, child age was also 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 were no significant differences in parental responsivity based on parent sex, there were differences in behavior management 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.

Overall, these findings suggest that parents of young children with FXS could benefit from interventions focused on increasing levels of responsive behaviors, especially given the association between child age and parental responsivity. 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 parent-implemented language interventions 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; 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 two and six years) and their mothers, mothers increased their use of responsive strategies, including comments and prompts for child communication (e.g., requests for verbal compliance). Moreover, the children in this study showed increases in their prompted communication acts (McDuffie, Oakes, et al., 2016).

Other studies of a parent-implemented language intervention 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, the measures of child language are from the same interactions being used to ascertain levels of parental responsivity. Future studies should incorporate additional external or distal measures of child language. 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 characteristics (e.g., ASD symptoms) 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 parent-implemented language interventions in families of children with FXS to increase parents' use of responsive strategies that target improvements in child communication.

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