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Parenting Young Children with and without Fragile X Syndrome

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Abstract

The purpose of this study was to examine maternal parenting styles across age-matched siblings using a within-family design, in which one child has Fragile X syndrome. Thirteen families participated; children were aged 16 to 71 months. Mothers completed several videotaped activities with each child separately as well as an interview. Mothers used a consistent, responsive style with both children, using the same degree of positive affect and warmth. Differences included using more behavior management strategies with the child with Fragile X and a conversational style of interaction with the sibling. Differences in approaches suggest the mothers adapted to the developmental differences between the children. The interview data supported these findings; mothers were aware of the changes made to accommodate the developmental differences.

Keywords

maternal parenting style; Fragile X syndrome; differential parenting; maternal responsivity

Parenting involves many complex processes and relationships sometimes leading to differential parenting of siblings within the same family unit (Rivers & Stoneman, 2008). Add to the mix a child with a developmental disability (e.g., fragile X syndrome, autism, Down syndrome), and the complexities of the family system and need for differential parenting may increase. The purpose of this exploratory study is to examine maternal parenting style with a specific focus on differential parenting using a multi-method approach. Our measurement of differential parenting is that of maternal responsivity. We selected this construct of parenting given the rich history of the impact of maternal responsivity on child development in both children with typical development (Landry, Smith, Miller-Loncar, & Swank, 1998; Landry, Smith, Swank, Assel & Vellet, 2001) as well as children with an intellectual disability (Crawley & Spiker, 1983; Roach et al., 1998; Warren et al., 2010).

Differential parenting has been studied in families of children with typical development, as well as families in which one or more children have a developmental disability, and has been linked to adverse outcomes in children such as poorer adjustment and relationship quality in children who receive less positive and more negative treatment (Asbury, Dunn, Pike, & Plomin, 2003; Deater-Deckerd et al., 2001). Studies that have examined child

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outcomes in relation to differential parenting have found that it was more pronounced in families with children with difficult temperaments and typical development (Feinberg et al., 2000; Jenkins et al., 2003). In terms of developmental disabilities, researchers have noted a trend for increased differential parenting in families with a child with a disability, typically with the child with the disability being favored in various ways (e.g., more attention, and positive parenting; Corter et al., 1992; McHale & Pawletko, 1992; Quittner & Opipari, 1994; Rivers & Stoneman, 2008). Generally these studies have relied on parent and sibling self-reports, and have not included direct observations of parent-child interactions.

One aspect of parenting that has been extensively studied is maternal responsivity. This construct refers to a healthy, growth-producing relationship between mother and child characterized by warmth, nurturance, and stability as well as specific behaviors such as mother's responses contingent on child initiations. A mother who is highly responsive will often engage in a style of parenting that maintains her child's focus of attention, expands on her child's initiations, and relatively rarely redirects her child to a new topic unless necessary. This style has been positively correlated with enhanced child language development, as well as enhanced cognitive, emotional and social development (Landry et al., 1998; Landry et al., 2001). On the other hand, directive behaviors such as attempting to refocus the child away from the focus of their attention may be characterized as less responsive, and result in a negative correlation with language development (Hampson & Nelson, 1993; Tomasello & Farrar, 1986). Thus, given its central role in parenting, we selected maternal responsivity as a focus for this study.

Maternal responsivity can be measured at several levels, from a more holistic level (i.e., molar responsivity) to a more detailed examination of behavior-by-behavior turn taking within a parent-child dyad, which we label molecular responsivity. Molar responsivity refers to a mother's general affect and verbal interaction style of parenting, and is typically measured via rating scales that assess behavioral interactions as having positive affect, warmth, and flexibility (Kim & Mahoney, 2004, 2005; Landry et al., 2000). Mothers of children with developmental disabilities have been reported to have lower affect scores and to be less responsive in their interactions compared to mothers of children with typical development (Kim & Mahoney, 2004).

Molecular responsivity refers to a more contingent pattern of specific responses. At this level, mothers are responsive if they respond to and expand child initiations and maintain the child's focus of attention (Spiker et al., 2002; Warren & Brady, 2007). This construct, labeled in some studies as maternal responsivity, corresponds with maternal behaviors that enrich the child's language learning environment (Warren et al., 2010). A focus on maternal responsivity may include attention to the behavior management techniques that can facilitate or inhibit development. One example of such a behavioral management technique is the use of redirects, which are typically defined as maternal control of children's behavior and/or attention. This type of redirect has been reported to at least modestly impede children's cognitive and language development if it occurs at high rates (Farran, 2001; Marfo, 1992; Mahoney & Neville-Smith, 1996).

Recent research with children with developmental disabilities, in particular studies of children with fragile X syndrome (FXS), demonstrate the usefulness of examining maternal responsivity in relation to child outcomes. FXS is an X-linked single genetic disorder associated with a number of impairments including intellectual disability in the majority of males, and in some females (Hessl et al., 2009). This disability has associated phenotypic characteristics that may alter parenting style. These include gaze avoidance or atypical eye gaze, hypersensitivity to sensory input, social anxiety and shyness, perseveration and repetitive behaviors, stereotypical behavior, unintelligible speech, and problems with

conversational discourse (Abbeduto, Brady, & Kover, 2007; Bailey, Hatton, & Skinner, 1998; Sterling & Warren, 2008). In addition, it has been estimated that between 25% and 45% of children with FXS also fall on the autism continuum and may display particularly severe forms of the behaviors noted above (Bailey, Hatton, Mesibov, & Skinner, 2000; Bailey, Hatton, Tassone, Skinner, & Taylor, 2001; Rogers, Wehner, & Hagerman, 2001). These deficits are in addition to problems with social interaction that may be associated primarily with developmental delay and intellectual disability. Given the challenging behavioral phenotype associated with FXS, it seems that even a parent with the best intentions could have a difficult time employing and maintaining a highly responsive style with their child. Given these challenges, it is unknown whether or to what extent parents of a child with FXS adapt their style to their child's individual needs, or adopt a consistent parenting style across their children with and without FXS.

A study by Warren and colleagues (2010) demonstrated that early maternal responsivity in very young children with FXS predicted later language outcomes after controlling for several child variables. The authors measured maternal responsivity, behavior management behaviors and child language outcomes at 36 months. They used a detailed analysis of structured and unstructured interactions between mothers and their young children with FXS to produce a molecular style of coding for responsivity. After controlling for child developmental level and autism symptoms, maternal responsivity predicted child expressive and receptive language on a standardized test (i.e. the Mullen) as well as on two important conversation based variables (rate of different words and total communication). In other words the more responsive a mother's style of parenting, the better the child language outcomes for these young children with FXS.

In the present study, given that the phenotype of fragile X includes challenging behaviors that could impede a mother's ability to maintain a responsive style of parenting (e.g., delayed language and cognition), we were interested in whether mothers are consistent between children in terms of their parenting style. It could be, for instance, that the mother continuously adapts her parenting style to be aligned with the developmental needs of the specific child, thereby exhibiting different parenting styles as a reflection of her assessment of what the child needs. To answer this question, we used a within-family design to directly compare a mother's parenting style with her child with FXS and her child with typical development.

We used several different measurement techniques, including an examination of responsivity and behavior management at both the molecular and molar levels, as well as mothers' report from semi-structured interview data. Participants in this study were 13 mothers of young children with the full mutation of FXS who also had a child typical development. Mothers were asked to complete a number of structured and unstructured interactional activities with their children with FXS, and then separately with their children with typical development. We were specifically interested in determining differences in the styles that mothers employed with the two children.

Method

Participants

We utilized a within-family design for this study. Thirteen sibling pairs (children with FXS and siblings with typical development) and their mothers from across the United States participated in the study. Recruitment efforts included advertising at national conventions, use of a national research registry housed at the University of North Carolina-Chapel Hill, networking with FXS family support groups, and advertising via an FXS parent listserv. Children with FXS were between 16 and 70 months of age (M = 46 months; see table 1 for

more information on the participants). In an attempt to control for age and, therefore, history of interaction with the mother, we used a chronological age match within each sibling pair. It was not possible for us to create a developmental match between the children. To get a chronological age match, we first completed the assessment with whichever child in the sibling pair was oldest. For 7 of the sibling pairs, this was the child with typical development; in 5 of the sibling pairs the child with FXS was the older child, and the last pair involved fraternal twins. Seven of the pairs were same gender (male-male), and 6 were male-female; all of the children with FXS were males. We waited until the younger sibling was the same chronological age to do the assessment (within a one month window). The average difference in age between the siblings at the time of the assessment was just 2 weeks.

Five of the siblings with typical development had not been tested for FXS, one child was a confirmed carrier, and seven children were not carriers and did not have the full mutation of FXS. Given that five children had not been tested for FXS, we completed the Peabody Picture Vocabulary Test third edition with each of the siblings to ensure the children had typical vocabulary (PPVT-III; Dunn & Dunn, 1997; see below for more information on the test and scoring). Eleven of the siblings completed the test. One child refused to complete the test, and testing was not completed for the other child due to time constraints; however, both of these children had been tested and were not carriers and did not have the full mutation. The mean PPVT-III score for the siblings was 110, with a range of 94 to 123, all within the range for typical development (see table 1 for more information on the mothers, children with FXS, and siblings). Three of the siblings, families 9, 10, and 13, were too young to complete the PPVT at the time of the assessment. We completed the PPVT with these children at a later date when they were within the appropriate age range for testing (minimum 2 years, 6 months).

We completed the Mullen Scales of Early Learning (MSEL; Mullen, 1995) with each boys with the full mutation in order to have a measure of developmental level. The boys in this study demonstrated a floor effect on the test, with a mean composite score of 51.2 (see table 1).

Molecular and Molar Coding

Molecular coding—Four different interactional contexts were videotaped and later coded to obtain the performance measures. Research assistants taped five minutes of each of the following contexts in the dyads' homes: 1) reading a book together, 2) making and eating a snack together, 3) unstructured play with toys of their choosing, and 4) a 30-minute sample of interaction in a "naturalistic" context. We judged that this range of contexts and the length of time samples in each were sufficient to provide a representative measure of the mother's behavior toward the child, and is in line with previous research on this topic (Roach et al., 1998; Warren et al., 2010). For the naturalistic context, mothers were instructed to conduct an everyday activity such as putting dishes away, folding clothes, or playing together. The three five-minute contexts and two 5-minute segments from the 30-minute "naturalistic" context were digitized for coding, yielding a total of 25 minutes of interaction. This amount of interaction coded is similar to that reported in other studies of maternal responsivity (Warren & Brady, 2007).

Graduate research assistants coded the digitized video files for mothers' communication and maternal responsivity using Noldus™ Observer software, version 5.1. Mothers' speech was coded on a behavior-by-behavior basis. The coding system used was adapted from Landry et al. (1998; 2001). All maternal behaviors and communication directed toward the child were coded using the codes defined in Table 2. When mothers' communication included several utterances in succession, the last utterance spoken to the child was coded based on the

assumption that the child's response would typically be anchored to the mother's final utterance.

Coding reliability—Graduate students were trained to identify and code behaviors to a training criterion of at least 80% agreement across three different samples before proceeding to code participant files. Once this criterion was met, two trained coders independently coded child and maternal behaviors for each observation file. Following the independent coding, the coders compared transcripts and resolved any disagreements through consensus. This process was implemented to ensure consistency across coders and over time. To determine the interjudge reliability for the variables analyzed — maternal responsivity and behavior management — intraclass correlation coefficients were calculated for each score (ICCs; Shrout & Fleiss, 1979). ICCs were calculated between the primary and reliability scores and between the scores arrived at by consensus and the primary independent codes. This procedure was used to determine whether the consensus coding procedure biased the data.

ICCs were high for child rate of communication behaviors, .98 calculated between primary and reliability and also .98 between primary and consensus coded data. ICCs were also high for maternal responsivity composite scores, .95 between primary and reliability ratings and . 97 between primary and consensus codes. For maternal behavior management consensus scores ICCs were similarly high, .88 between primary and reliability and .92 between primary and consensus codes. These strong correlations indicate that differences in the final behavior codes derived from consensus coding and those of primary coders had a very small effect on the reported performances of participants.

Molar coding—Mother-child dyads were instructed to participate in a 30-minute unstructured "naturalistic" interaction (described previously in the molecular coding section). During this context, mothers were instructed to conduct an everyday activity such as putting dishes away, folding clothes or playing together. The videotapes were then digitized and coded using a system adapted from Landry et al. (2000). Videotapes were scored on 3 different 10-minute intervals, and then the 3 intervals were averaged for a final composite score. Three maternal behaviors were coded (see Table 3 for definitions). The behaviors were rated on a scale of 1 to 5, with 1 representing nonexistent behaviors (i.e., positive affect scale, a rating of 1 corresponded with 0 smiles), and a score of 5 as the highest score.

Undergraduate and graduate students were trained on the rating scales. Every third file was reliability coded to ensure that reliability stayed at or above 80%. The mean percent agreement for positive affect was 94%, warmth 94%, and flexibility/responsiveness 92%. Reliability for the three scales combined was 93.3%.

Standardized and Descriptive Measures

The Mullen Scales of Early Learning (MSEL; Mullen, 1995)—This standardized developmental test for children ages 3 to 60 months is administered via direct observation and testing. It is comprised of the following subscales: Visual Reception, Fine Motor, Receptive Language, and Expressive Language. Each subscale yields a standardized score, and these scores are summed together to create an overall composite score. The standardized scores for the subtests are based on a mean of 50, while the overall composite score is based on a mean of 100, with a standard deviation of 15. The boys in this study had a mean composite score of 51.2; in other words, the boys were moderately to severely delayed. The subscale scores reported within the analyses are based on raw scores, while the reported composite score is the standardized score.

Peabody Picture Vocabulary Test Fourth Edition (PPVT-III; Dunn & Dunn, 1997)—This standardized test was used with the siblings only to ensure the children were developing typically. Participants are asked to point to a visual representation of a word spoken by the examiner. The PPVT-III is a standardized test; age equivalent scores can be calculated based on results. The mean score for this test is 100, with a standard deviation of 15. The siblings in this study had a mean score of 110, with a range from 94 to 123. None of the siblings were in the impaired range.

Wechsler Adult Intelligence Scale — third edition (WAIS-III; Weschler, et al., 1997)—This standardized measure was used for maternal IQ. The test is normed for adults between the ages of 16 to 89 years, and typically takes 60 to 90 minutes to complete. Full scale IQ scores are calculated from subtests. See table 1 for more information.

Maternal education—Mothers were asked to complete a basic demographic form indicating number of years of education completed. Each mother completed this at the time of the assessment.

Semi-structured Interviews

To provide more contextual information on mothers' descriptions of interactions with their children and to provide concrete examples of maternal responsivity and behavioral management strategies, we conducted semi-structured interviews before administering the quantitative assessments. These interviews elicited information on a number of domains (e.g., quality of life, impact of diagnosis), and included a section devoted to maternal responsivity. Interview questions elicited mothers' perceptions of their parenting styles, the strategies they used to communicate with their children and manage their behavior, and whether these styles and strategies were the same or different for the child with FXS and the typically developing sibling. The interviews lasted approximately 90 minutes, were digitally recorded and transcribed verbatim (for other articles based on these interviews, see Brady et al., 2006; Michie & Skinner, 2010; Raspberry & Skinner, 2011; Wheeler, Skinner, & Bailey, 2008). The second and third author conducted a content analysis (Bernard & Ryan, 1998; Miles & Huberman, 1993) of the interview data to determine the range and type of strategies mothers said they used, and to identify examples of behavioral management and maternal responsivity that corresponded to the constructs defined by the codes above.

Results

The purpose of this study was to examine and compare maternal parenting style across siblings in the same family. We used several different measurement techniques, including an examination of responsivity and behavior management at both the molecular and molar levels, as well as mothers' reports from interview data. Results are reported by each maternal behavior composite: the two molecular types of maternal behavior (maternal responsivity and behavior management), and molar responsivity. Accounts from the interviews are used to provide examples for the ways in which mothers reported interactions with their children that were related to these constructs.

For this study we used the same maternal-child interactional contexts and maternal coding system as used in a previous analysis on parenting style in mothers of children with FXS (see Warren et al., 2010). In Warren et al. (2010), we reported results on two components comprised of coded maternal behaviors based on a Principal Components Analysis (Gorsuch, 2003). The first component consisted of gesture use, request for verbal complies, comments and recodes (i.e., verbal interpretations of the child's communication act). We labeled this maternal responsivity. The second component included redirects, request for

behavioral complies and zaps (i.e., restrictions of child behavior, for example, “No, stop that.”). We labeled this component behavior management. The following results are broken down into these components, as well as a molar system of coding (i.e., molar responsivity).

We first ran correlations between the maternal and child variables and the composite scores from the quantitative data to do an initial evaluation of the relationships between the variables (see Table 4 for more information). There was a significant negative correlation between behavior management and the sibling’s developmental level. Additionally the correlation between the developmental level and behavior management of children with FXS approached significance ($p = .06$). We then completed paired samples t tests to compare parenting style within the sibling pairs. Given the small sample size, we calculated effect sizes to complement the findings. For each type of responsivity we report both quantitative and qualitative findings.

Maternal Responsivity

Mothers used comments at a significantly higher rate with their children with typical development, $t(12) = -3.03$, $p = .01$, $d = -.92$ (TD: $M = 3.33$, $SD = 1.00$, FXS: $M = 2.60$, $SD = .52$). The other three maternal behaviors comprising this component (e.g., request for verbal complies, recodes, and gestures) did not yield significant differences, although there was a medium effect size for rate of request for verbal complies, $t(12) = -1.40$, $p = .19$, $d = -.51$ (TD: $M = 2.78$, $SD = 1.04$, FXS: $M = 2.16$, $SD = 1.37$).

Behavior Management

There was a significant difference for rate of redirects, $t(12) = 2.55$, $p = .025$, $d = .870$. The mothers were using redirects at a rate of .108 times per minute for their child with typical development, and .251 for their child with FXS. All mothers reported using redirection with their child with FXS, and 10 mothers said they used it with both children but employed it more frequently with the child with FXS. Mothers used redirection to guide children’s attention to an intended activity, to prevent children from becoming upset in situations that had previously caused them to become upset, and to prevent an undesirable behavior. When mothers compared their children, they saw the behaviors of the child with FXS as being less intentional than the same behaviors of their children who were typically developing. For example, one mother explained that when her son with FXS hit his brother, it was rarely on purpose but rather a result of him having “low tone” and problems with his “proprio-receptors.” She said that he only intentionally struck his brother when the brother hit or goaded him. She described how she differentially treats the two boys, saying her son who is typically developing “understands me… [knows] what my expectations are, and so I don’t feel like I have to put as much effort into him,” but that she “goes easy” on her son with FXS. Another difference is that most mothers continued to use redirection for the child with FXS, but not with the sibling as they aged.

We did not find statistically significant differences between the sibling pairs in the other two maternal behaviors for behavior management; however, we did find medium effect sizes for both behaviors: rate of zaps, $t(12) = 1.57$, $p = .14$, $d = .74$ (TD: $M = .24$, $SD = .15$, FXS: $M = .42$, $SD = .31$); and rate of behavioral complies $t(12) = 1.77$, $p = .10$, $d = .68$ (TD: $M = 1.65$, $SD = .50$, FXS: $M = 2.07$, $SD = .72$). We did not ask in interviews specifically about the use of “zaps,” but all mothers described examples that fit this category. One mother, when asked what she did when she disciplined her child with FXS, replied, “I say ‘I want you to go up to your room, and I want you to lay on the bed, and you come back down once you’ve rested.’ …We’ll [also] put him in time out from time to time.” When asked whether she still put the sibling in time out, she said, “He does not need a time out anymore. He’s really just lovely right now…usually you can just reason with him.” Similar to using

redirection, 11 of the 13 mothers reported continuing to use time outs with their children with FXS while discontinuing this strategy with the siblings as they matured. Though children with FXS and their siblings received time outs at some point, some mothers reported that the siblings matured out of time outs while none of the children with FXS had.

Eleven of the mothers also reported using requests for behavioral comply with their children with FXS, such as "Get in the bath, take your clothes off, put the clothes in the hamper." These requests for behavioral comply most often involved specific instruction that broke down an activity to enhance the child's comprehension.

Molar Responsivity

There was a statistically significant difference in terms of the flexibility/responsiveness code in the sample, $t(12) = -2.58$, $p = .02$, $d = -.93$ (TD: $M = 3.97$, $SD = .59$, FXS: $M = 3.40$, $SD = .62$). Mothers demonstrated less flexibility and responsiveness toward their children with FXS compared to their children with typical development. The other two maternal behaviors of warmth and positive affect did not yield significant differences, and the effect sizes were small or nonexistent. In other words, the mothers were engaging in the same levels of warmth in their interactions with both children, and smiling at both children the same amount.

Interview data supported this finding in that nearly all mothers described how they encouraged their children's desirable behavior, and did so in similar ways, especially when both siblings were at younger ages. They used verbal praise, smiles, and clapping. One mother said for her son with FXS, she used: "Verbal praise and you can tell cause you can say, 'Good job' and he'll go (acts excited). He gets all excited and happy." She reported she used the same tactics with her child who is typically developing.

Despite mothers' frequent statements that they encouraged their children in the same ways, they also noted that their children with FXS preferred more or less effusive praise than their siblings who were typically developing, or had more or less preference for physical contact (e.g., hugs or 'high-fives'). They stated that they tempered their praise accordingly for each child. They also noted that the ways they encouraged their children changed as children with typical development often required more than praise at earlier ages than their children with FXS. This would suggest that the mothers might have been responding to the child's developmental level, irrespective of their age. Seven mothers reported using a reward system such as accumulation of points or tokens towards privileges or items with both children, but four said they did not find this as effective for the child with FXS who was less able to comprehend a reward granted for accumulation of achievements over time. In general, all mothers described their most common strategy with the child with FXS to be positive consequences (e.g., praise) given at the time of 'good' behavior. One mother explained:

> Well, verbal positive reinforcement is still huge for him (son with FXS) so I do a lot of praising, a lot of hugging. Letting him play with something he wanted to play with or a special activity I know that he would want to do… I still give a lot of verbal reinforcement and hugs and special activities to my other kids but then there's another level. Like if they do chores, then they get money so there's a little bit higher level of reward, or if they get straight A's on their report card, then they get a King's Island season pass… that's the difference with (son with FXS)— it's more like a short-term reward. Like this is like an instant reward. For (siblings) it could be something over several months and get a reward.

Discussion

The quantitative analysis and interview data both indicate that in general, mothers used the same types of parenting strategies with both children. The general picture from this study is a positive one: mothers used lots of praise and warmth with both children (molar responsivity) regardless of their differences in developmental level; the differences in parenting for both molecular and molar codes focused on maternal behaviors central to either diffusing problematic child behaviors or in their use of a more conversational style of language with their children with typical development. In other words, the mothers were not exhibiting high levels of differential parenting between siblings, but rather they were employing a responsive style of parenting with both of their children that likely accommodated appropriately for the developmental differences in the children.

In terms of the behavior management factor we found that the rate of redirects was the only significantly different behavior. Mothers redirected their children with FXS significantly more often compared to their children with typical development. The mothers commonly used redirects as a way to manage undesirable behavior. All mothers described redirection as a strategy they applied to both intentional and unintentional undesirable behaviors. According to interview data, mothers often used redirections at later chronological ages with their child with FXS compared to their child with typical development, again possibly reflecting the developmental differences. This was especially the case with “time out,” which is not surprising given that children with FXS often display problematic behaviors, including aggression and tantrums, through later childhood, whereas most children with typical development cease using these behaviors at an earlier age. Some research (Farran, 2001; Marfo, 1992) has indicated that redirecting a child’s attention is negatively associated with child language outcomes. However, despite the statistically significant difference in redirects by the mothers, the actual rates of redirects were not frequent as compared with other maternal behaviors such as comments and gestures, with a rate of .25 instances per minute for the mothers with their child with FXS, and .108 for their children with typical development. The mothers noted in their interviews that redirects were used to manage undesirable behaviors, and to guide them to intended activities, and that this was necessary more frequently with the children with FXS. Perhaps this infrequent use of redirects is not as negative in FXS; future work on this topic should continue to disentangle this result.

Our maternal responsivity factor indicated that mothers were using comments more frequently with their child with typical development compared to their child with FXS, and interactions between the mother and the typical sibling were more likely to be conversational overall. This again is likely a reflection of the developmental differences between the age-matched children (see table 1 for more information on children’s developmental level).

The majority of the sibling pairs were between the ages of 2 and 5 years of age at the time of data collection. In families where a child has an intellectual disability, maternal responsivity studies have indicated that particularly in children under the age of 5, mothers bear more of the burden of the interaction (Crawley & Spiker, 1983; Tannock, 1988). This appears to be the case in this study.

For other maternal responsivity variables, there were no differences in terms of gesture use or recoding of child speech, which was unexpected given the differences in the children’s developmental levels. One general expectation mothers noted in interviews for their children with FXS was an improvement of the child’s communication skills. Recoding speech is a classic behavior used to help expand not only child vocabulary, but also phonology, semantics, and grammar (Ebbels, 2008; Nicholas, Lightbown, & Spada, 2001). However,

mothers were not using this strategy with a greater frequency with the children with FXS. In fact, although there was only a small effect size, the means indicate that the mothers were using recodes more often with their children with typical development. It is important to remember that the children with typical development were communicating significantly more, thus providing more opportunities for recodes. It could also be the case that using recoded speech in their verbal interactions with their young children became common practice, regardless of need. Two of the molar responsivity codes, positive affect and warmth, did not yield significant differences, nor notable effect sizes. It seems the mothers were showing physical and verbal warmth and praise at the same rates for both groups of children. Mothers noted in their interviews that they often used verbal praise for both of their children. Verbal praise is one component of warmth. Perhaps the amount of smiling a mother does, and her overall level of warmth is more entrenched in her identity as a parent and in her parenting style, and therefore does not change between children.

The flexibility/responsiveness code was significantly different, with mothers showing less flexibility in their interactions with their children with FXS. It is important to note that while there was a significant difference in flexibility, the means were not so great as to indicate that the mothers were completely rigid with their children with FXS, or entirely flexible with their child with typical development.

This study has both strengths and limitations. The within-family design allowed us to examine parenting style within the same family unit, providing a direct comparison of how parenting style differs with children at the same chronological age and whether their style was consistent within the family. Due to time and resource constraints, we had to employ a chronological age match instead of a developmental age match. Had we been able to match for development, we may have found even fewer differences in the mothers' parenting styles. Another limitation is the broad age range of the children (16 months to 70 months). Ideally, we would have selected a narrower age range but these children are difficult to identify at the younger ages. Also given our small sample size, we were not able to match the children on gender, or the mothers on variables known to impact responsivity such as education level. Our mothers were well educated, and the findings could be different with a broader educational range; additionally the fact that this was a self-selected sample could have biased the results. Our participants were connected within a broader community of individuals with FXS, and were most likely more informed on the behavioral phenotype compared to the general population.

Despite the small sample size, and exploratory nature of the study, we found a clear pattern of results. The mothers in this sample were not exhibiting high levels of differential parenting between siblings, but rather they were employing a responsive style of parenting with both of their children that likely accommodated appropriately for the developmental differences in the children. There are a number of future directions from these findings. Given the young ages of our participants, we did not include a self-report measure for the siblings. This has been done in previous research and would be interesting in an older sample of children. Additionally, we did not use a measure of autism in our study. Exploring the impact of autism on the family system in FXS is an important and interesting next step in this line of research.

We believe this study has at least one important clinical implication. There is a body of research indicating that responsive parenting styles can be acquired by parents of children with developmental disabilities with relatively modest amounts of parent training (for review see Warren & Brady, 2007). For the mothers in this study, the molar measures of responsivity such as positive affect and warmth were consistent between siblings. However, mothers were using redirects more often with their children with FXS, although at a low

rate. Given the importance of responsivity on later language and cognitive development, the mothers could benefit from training on more responsive parenting.

Mothers were generally utilizing a responsive style of parenting with both of their children and broad indicators of differential parenting were not observed in this sample. Mothers' reports of their parenting style in interviews aligned with their coded interactional behaviors. In terms of molar behaviors, the mothers displayed the same degree of positive affect and warmth in their interactions with both children, and they provided a rich language learning environment for both children, including the use of gestures and recodes.

Differences in parenting were observed for behaviors used to manage problematic behavior, such as redirecting the child's attention. In interviews, mothers noted that these strategies were used to manage challenging behaviors in their children, and that they had to use these strategies more often with their children with FXS. To summarize, in this sample of mothers of children with FXS, parenting style was relatively consistent regardless of disability, and where it did differ may reflect differences in the child's behavior. This finding mirrors what is found in the literature on children with typical development (Feinberg et al., 2000; Jenkins et al., 2003). Although the sample size was small, this study suggests that mothers are generally able to maintain their overall parenting style regardless of the presence of FXS, while being in tune with the child's needs and making adjustments when necessary.

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Table 1

Descriptive Information for Mothers, Children with FXS, and Siblings

	Maternal IQ	Maternal Education Level		Chronological Age in months	Developmental Level Test Scores[a]	Gender
Family 1	128	19	Child with FXS	62	53	M
			Sibling	61	94	M
Family 2	108	17	Child with FXS	35	49	M
			Sibling	35	121	F
Family 3	110	18	Child with FXS	70	55	M
			Sibling	70	117	F
Family 4	111	13	Child with FXS	40	49	M
			Sibling	41	*	M
Family 5	125	18	Child with FXS	55	49	M
			Sibling	55	117	M
Family 6	123	15	Child with FXS	67	49	M
			Sibling	68	118	M
Family 7	122	17	Child with FXS	66	52	M
			Sibling	66	101	F
Family 8	119	16	Child with FXS	36	50	M
			Sibling	36	95	F
Family 9	111	15	Child with FXS	22	54	M
			Sibling	22	111	M
Family 10	86	14	Child with FXS	16	50	M
			Sibling	16	91	F
Family 11	121	14	Child with FXS	36	49	M
			Sibling	37	123	F
Family 12	99	16	Child with FXS	71	49	M
			Sibling	69	104	M
Family 13	101	16	Child with FXS	19	58	M
			Sibling	19	*	M

Note. Rows represent individual family units: mother, child with FXS, and typically developing sibling.

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[a]Developmental level: The Mullen Scales of Early Learning composite score was used for the children with FXS; the siblings were given the Peabody Picture Vocabulary Test (PPVT-III; citation). Standardized scores are presented for the participants.

*Missing data

Table 2

Definition of Molecular Codes

Behavior:	Definition:	Example:
Maternal Responsivity		
Gestures	Sign language, gestures (“come here,” “stop,” “no”), tapping, clapping, or knocking, etc.	Mother points to the book and says “Do you want to read this?”
Request for Verbal Comply	Question/statement aimed at getting a verbal response	Mother says, “Say ___,” or “Huh” or “ok” at the end of a comment.
Comment	All comments	Praise in reaction to something the child has done.
Recode	Verbal interpretation of child’s communication act	Child says “ba” and mother says “Do you want your ball?”
Behavior Management		
Request for Behavioral Comply	Directives to which the child can comply behaviorally	Mother says, “Push this one,” or “I want you to do it.”
Redirect	Mother references new object when child is actively attending to another object	Child is playing with a toy and mother says, “What else do you want to play with?”
Zap	Restricting child’s behavior in some way – not always negative	Mother says, “No stop that,” “Don’t touch that.”

Table 3

Definition of Molar Codes

Behavior:	Definition:	Example:
Molar Responsivity		
Display of Positive Affect	Number of times a mother smiles directly at child	Smile must be directed at the child; mother's teeth must be visible
Warmth	Mother responds to child's verbal and nonverbal communication attempts; praise; enthusiasm	Mother who is involved and who does not have a negative tone would have a higher score
Flexibility/Responsiveness	Mother's agenda vs. child's; sensitivity to child's cues; amount of involvement with child	Mother who is highly responsiveness will be involved, but also patient for child's abilities.

Table 4

Correlations between Molecular and Molar Responsivity with Maternal and Child Variables

	Maternal IQ	Maternal Education	Mullen	PPVT
Maternal Responsivity	.38	.30	.47	.19
Behavior Management	.17	−.42	−.54^{+}	−.70*
Molar Responsivity	.11	.06	−.06	.05

Note. Significance values:

$^{**}p < .01$;

$^{*}p < .05$;

$^{+}p < .06$