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Influences on maternal responsivity in mothers of children with fragile X syndrome

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Abstract

This study investigated the influence of maternal and child variables on the maternal responsivity of 55 mothers with young children with fragile X syndrome. Data included video observations of maternal-child interactions in four different contexts, standardized assessments with the children, and standardized questionnaires for the mothers. The video observations were coded for child communication acts; maternal responsivity was coded at two levels: a more general measure and a behavior-by-behavior measure. Results indicated that child developmental level and language ability strongly influenced behavior-by-behavior responsivity, while maternal IQ was the strongest predictor of both general and behavior-by-behavior responsivity, after controlling for child developmental level.

Maternal responsivity refers to a healthy, growth-producing relationship characterized by warmth, nurturance, and stability as well as specific behaviors such as responses contingent to individual child initiations. Maternal responsivity has been shown to have a cumulative impact on children's cognitive, emotional, and language development (Landry, Smith, Miller-Loncar, & Swank, 1998; Landry, Smith, Swank, Assel & Vellet, 2001). 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 rarely redirects her child to a new topic. On the other hand, high rates of directiveness, which are typically defined as maternal control of children's behavior and/or attention, may impede children's cognitive and language development (Farran, 2001; Marfo, 1992; Mahoney & Neville-Smith, 1996).

Warren et al. (2010) investigated the relationship between early maternal responsivity and later child communication outcomes in young children with fragile X syndrome (FXS). Data

were obtained from 55 mother-child dyads over a 36 month period. Performance data were obtained from video observations of 4 different contexts. These were coded for child communication behaviors, maternal responsivity, and maternal behavior management. Results demonstrated that higher levels of maternal responsivity predicted later receptive and expressive language scores on standardized tests, as well as rate of number of different words used by the child, and the child's total communication (including both verbal and nonverbal communication acts). The authors controlled for child behaviors including autism symptomology for all four variables, and overall child developmental level as indexed by the Mullen Scales of Early Learning (MSEL; Mullen, 1995) for two of the child outcomes (child's total communication and number of different words).

Given the importance of maternal responsivity to child development, it is in turn important to determine variables that influence maternal responsivity. Previous research has identified a number of parent and child variables that may influence a mother's ability to employ a highly responsive parenting style including parental emotional state (e.g., depression), beliefs and values (e.g., "spare the rod spoil the child"), and maternal education level, as well as general child variables such as the child's temperament and developmental level (Shapiro, Blacher & Lopez, 1998). The present paper extends the Warren et al. (2010) study by examining the influence of several maternal and child variables on maternal responsivity using the same 55 mothers and children with FXS.

Responsivity can be defined and measured in different ways. Warren and Brady (2007) described three levels: general responsivity, molar responsivity, and molecular responsivity. At the broadest level, general responsivity refers to responding to a child's most basic needs (e.g. food, shelter, education, etc). Molar responsivity typically refers to a mother's general affect and style of parenting, and has typically been measured via rating scales (Kim & Mahoney, 2004, 2005; Landry, Smith, Swank, & Miller-Loncar, 2000). Dimensions such as "positive affect", "warmth", and "flexibility" in interactions are examples of molar responsivity measures (e.g., Landry et al., 2001). Mothers of children with developmental disabilities have been reported to have lower affect scores and to be less responsive at a molar level of interaction compared to mothers of children with typical development (Kim & Mahoney, 2004).

Molecular responsivity refers to a behavior-by-behavior level of interaction between caretaker and child. At the molecular level mothers may be considered highly responsive if they respond to and expand a relatively large proportion of child initiations, and maintain the child's focus of attention (Warren & Brady, 2007). A number of studies have investigated molecular measures of responsivity with children with developmental disabilities including Down syndrome (Crawley & Spiker, 1983; Roach et al., 1998; Slonims & McConachie, 2006; Tannock, 1988). These studies indicate that mothers of children with disabilities tend to be relatively more directive and carry the burden of the interaction.

Maternal responsivity (both molar and molecular) represents a complex, dyadic, interaction involving both child and maternal variables. Certain maternal variables, such as low educational attainment (i.e., less than a high school education), mild intellectual disability, substance abuse, and depression have been shown to have a significant impact on a mother's

ability to maintain a highly responsive style of parenting in children with typical development (Hooper, Burchinal, Roberts, Zeisel, & Neebe, 1998; Miller, Heysek, Whitman, & Borkowski, 1996; Osofsky & Thompson, 2000; Rutter & Quinton, 1984). The biological mothers of children with FXS are themselves premutation carriers of FXS (a small portion may have the full mutation as well), and therefore are at risk for a range of subtle to severe cognitive or emotional problems that may hinder their interactions with their children. These risks include cognitive deficits in attention, verbal memory, and executive function (Freund, Reiss, & Abrams, 1993; Sobesky, Hull, & Hagerman, 1994); additionally women with the premutation of FXS are more prone to depression, social anxiety, and may be more affectively labile (Hagerman, 2002; Mazzocco, 2000; Sobesky, et al., 1994; Thompson et al., 1996). These risk factors have been associated with lower maternal responsivity (Goldsmith & Rogoff, 1995; Osofsky & Thompson, 2000); therefore it is important to examine the impact of these factors on maternal responsivity in this group of women.

Depression and social anxiety have been examined in mothers of children with developmental disabilities, including autism and FXS. Researchers have found lower rates of well being in mothers of children with autism, and have linked this to the higher rates of maladaptive behavior (Koegel et al., 1992; Seltzer et al., 2000). In terms of FXS, studies have found higher lifetime depression levels in premutation carriers (56%), compared to the general population (10-12%, NIMH, 2003; Abbeduto et al., 2004; Wheeler, Hatton, Reichardt, & Bailey, 2007). However, the mothers with the premutation did not report higher levels of depression compared to mothers of children with other developmental disabilities (Abbeduto et al., 2004). Much like the autism literature, the most consistent predictor of maternal stress was the severity of child problem behaviors in this group of mothers.

Different maternal traits can influence responsivity, and this interactional process is influenced by the behavior of both the mother and child. A mother with the best intentions can still have difficulty employing and maintaining a highly responsive style with a child with a developmental disability (Stormont, 2001). Children with FXS may have any of a number of phenotypic characteristics such as hyperactivity, gaze avoidance, unintelligible speech, and perseverative and stereotypic behaviors, and passivity (Abbeduto, Brady, & Kover, 2007; Bailey, Hatton, & Skinner, 1998; Sterling & Warren, 2008). These behaviors could make a highly responsive interaction difficult, which over time may cause caregivers to become less responsive even to appropriate initiations (Murphy & Abbeduto, 2005).

It is estimated that approximately 15%–25% of individuals (mostly males but some females) with FXS meet the diagnostic criteria for autism (Clifford, Dissanayake, et al., 2007; Rogers, Wehner, & Hagerman, 2001). Furthermore, regardless of co-diagnosis, 50%–90% of males with FXS are reported to show some of the symptoms of autism, including self-injurious behaviors, stereotypic movements (i.e., rocking, hand flapping), perseverative speech, tactile defensiveness, and poor eye contact (Bailey et al., 1998; Clifford et al., 2007; Feinstein & Reiss, 1998). Males with both FXS and autism typically have more severe language and social impairments, as well as lower IQ scores compared with children with FXS without autism (Bailey et al., 1998, 2000). In light of the fact that severity of

behavioral problems is a predictor of maternal well being, autistic symptoms are an important variable to examine in determining the impact of the child's behavior on maternal behaviors.

The present paper is an extension of the Warren et al. (2010) study, which reported the impact of early maternal responsivity on four child language outcomes. The present investigation focuses on the child and maternal variables that in turn influence that measurement of maternal responsivity in the same sample of 55 dyads. The maternal variables of interest include maternal education, maternal IQ, maternal stress (measured by Parenting Stress Index; Abidin, 1995), and depression (Beck Depression Inventory; Beck et al., 1996). The child variables of interest are child development as measured by the Mullen Scales of Early Learning (Mullen, 1995), autism symptomology as measured by the Childhood Autism Rating Scale (CARS; Schopler, Reichler & Renner, 1988), child temperament measured by parent report (Difficult Child subscale on the Parenting Stress Index; Abidin, 1995), and a coded measure for rate of child communication. We examined the influence of these variables on two levels of maternal responsivity: molar and molecular.

Method

Participants

Fifty-five children with FXS between 11 and 48 months of age and their biological mothers were recruited from across the United States. 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 list serve.

This was a sample of convenience due to the fact that FXS is a rare disorder with few symptoms at birth and children are often not identified until age 3 or older. Given our national recruitment efforts we were able to identify families who had very young children with FXS, and were therefore identified before the national average. Although desirable, we were not able to tightly control either age of entry into the study or other potentially important SES variables. Nevertheless, we were able to get a reasonable amount of variability on factors such as maternal education level and family income, but less diversity than desired in terms of racial composition. The sample included 44 boys and 11 girls with the full mutation of FXS. Table 1 provides descriptive information on the child participants. Composite scores from the Mullen Scale of Early Learning (Mullen, 1995) provide an estimated level of development. The Childhood Autism Rating Scale (CARS; Schopler et al., 1988) provides a well validated measure of autistic symptoms (Rellini, Tortolani, Trillo, Carbone & Montecchi, 2004). A score in the range of 30-36 on the CARS is considered to be in the mild to moderate range for autism. A score above 36 is considered an indicator of severe autism. We did not use the CARS score as a diagnostic indicator, but simply as a measure of the presence of autistic behavior for each child participant during the time that we observed them.

Three of the mothers had the full mutation of FXS, and the other 52 mothers were premutation carriers. We included the three mothers with the full mutation in the study since

they represent a small but very high risk segment of mothers of children with FXS, thus making our sample more representative. Each mother completed the Wechsler Adult Intelligence Scale—third edition (Wechsler et al., 1997). Basic demographic information was collected, including race and ethnicity, income, and maternal education. The full sample of mothers represented a wide range of incomes with 23% (14 mothers) of the sample qualifying as “low income”. Eighty-three percent of the sample were married and 88% were Caucasian. Education level ranged from 6 parents (11% of sample) with a high school education or less to 53% who had graduated from college. The three mothers with the full mutation varied in terms of education level. The mother with the full mutation with the lowest IQ completed high school, the mother with an IQ of 89 completed 3 years post high school and the mother with the IQ of 103 completed 4 years post high school (see table 2 for more detailed information).

Measures

Data were collected on each child with FXS and his/her biological mother including direct measures of maternal responsivity and child communication observed during mother-child interactions in four different contexts.

Molecular Coding—Four different interactional contexts were videotaped and later coded to obtain the performance measures in this study. Five minutes of each of the following contexts were videotaped in the dyads’ homes: reading a book, making and eating a snack, unstructured play with a set of toys appropriate for age and developmental level, and a 30 minute sample in a “naturalistic” context, all within the course of the visit. The first three contexts were videotaped in succession, and there was generally a break before the 30-minute sample. We judged that this range of contexts and the length of the time samples were sufficient to give us a representative measure of the child’s language as well as the mother’s behavior to the child. During the naturalistic context, parents 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. The two 5-minute segments from the 30-minute sample were minutes 5 to 10, and 20 to 25 of the total 30 minutes. This amount of coded interaction is similar to that reported in other studies of maternal responsivity (Warren & Brady, 2007; Warren et al., 2010).

The digitized video files were coded using Noldus™ Observer software, version 5.1. The child measure coded was rate of total communication observed during the interaction. Rate of total communication was obtained by counting the number of all communication acts coded and dividing this number by the total time. Communication acts included words, signs, symbols, intentional gestures or intentional vocalizations. Each videotaped observation file was also coded for maternal responsivity using the Noldus™ software. The mother’s 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 was coded using the codes listed in Table 3. 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—Both undergraduate and graduate students were trained to identify and code the behaviors to a training criterion of at least 80% agreement across three different samples before being allowed to code participant files from the current investigation. Once this criterion was met, two trained coders independently coded child and maternal behaviors for each observation file. Following the independent coding, transcripts were compared and any disagreements were resolved 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, maternal behavior regulation and two proximal child communication outcomes - intraclass correlation coefficients (ICCs; using the absolute agreement and single measure values) were calculated for each score (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 (see table 4). 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 interaction. 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 10-minute intervals, and the 3 intervals were averaged for a final composite score. Three maternal behaviors were coded (see table 5 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 no observed 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 in order to ensure that reliability stayed at or above 80%. The mean percent agreement for positive affect was 93%, warmth 92%, and flexibility/responsiveness 92%. Reliability for the three scales combined was 92.3%.

Child standardized 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. We used an overall raw score as our composite variable for child development based on the four subscales.

Childhood Autism Rating Scale (CARS; Schopler, Reichler & Renner, 1988)—

This 15-item measure provides a general rating of autistic behavior. Each item is rated by the examiner on a score from 1 (within normal limits for age or developmental level) to 4 (severely abnormal for age or developmental level). The total score can be used as a continuum of autistic behavior. Scores on the measure can be interpreted as non-autistic (total score of 15-29.5), mildly or moderately autistic (total score of 30-36.5), and severely autistic (total score of 37 or higher). The CARS was administered by two trained examiners following the assessment. For scoring purposes, the mildly-moderately autistic and severely autistic scores were collapsed. As a result a dichotomous variable was created: non-autistic (scores from 15 to 29.5) and autistic (any score above a 30).

Parenting Stress Index (PSI; Abidin, 1995)—The PSI is a standardized questionnaire designed to identify dysfunctional parenting by examining both parent and child constructs. The PSI consists of 120 items, and generally takes 20 to 30 minutes to complete. Mothers were mailed the form prior to the assessment in order to allow plenty of time for completion of the questionnaire, and forms were collected at the assessment. Three of the mothers did not complete the form. We used the Difficult Child standardized score as a child predictor in our analyses.

Maternal standardized and unstandardized measures

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.

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. The mean IQ for the full sample of mothers was 107 and the range of scores, 55-130, was substantial. The three mothers with full mutation FXS had IQ scores of 55, 89, and 103.

Parenting Stress Index (PSI; Abidin, 1995)—For the purposes of this paper, we used the Parental Distress standardized score in the maternal analyses. Seventeen of the fifty-five moms in this sample reported clinically significant levels of stress.

Beck Depression Inventory-II (BDI-II; Beck, Steer, & Brown, 1996)—The BDI-II is a self-report measure of current depression (within a two week period). It consists of 21 items and generally takes 5 to 10 minutes to complete. It is designed for use in both the clinical and general population and is based on criteria from the DSM-IV. Mothers were given the forms at the time of the visit. Three mothers did not complete this form. Five

mothers reported scores in the range of mild depression, 2 in the range of moderate depression, and one in the severe range.

Results

Principal components analysis

The focus in the Warren et al. (2010) paper was on the influence of early maternal behavior (at the molecular level) on subsequent child language outcomes. A preliminary analysis revealed a substantial level of correlation among the seven maternal behavior codes for young children aged 19 to 37 months. Therefore the authors conducted a principal components analysis (Gorsuch, 2003), which reduced the seven maternal codes to two principal components for observations made within that age range. The first component included gesture use, requests for verbal complies, comments, and recodes. The authors labeled this maternal responsivity. The second component consisted of redirects, requests for behavioral complies and zaps (restrictions of child behavior, e.g., “No, stop that.”). This was labeled behavior management. In order to combine scores that were quite varied in scale into a single component score, the authors computed z-scores for each indicator and then averaged the z-scores across the scores within the factor for each participant. These same molecular factors, maternal responsivity and behavior management, were explored in the present paper. In addition, we created a composite consisting of the molar variables described earlier. The composite was made up of three subscales: display of positive affect, warmth, and flexibility/responsiveness. Z-scores were created for each subscale, and the composite consisted of the mean of the z-scores. This factor was labeled molar responsivity. We ran correlations between our factors. Molar responsivity was significantly correlated with Maternal Responsivity, $r = .35, p < .01$; however it was not significantly correlated with Behavior Management, $r = .06, p > .05$.

Regression analyses

Multiple linear regression analysis was used to model the effects of child and maternal predictors on maternal behaviors. The dependent variables were the maternal responsivity, maternal behavior management, and the molar responsivity composites. The independent variables used as child predictors were: 1) Mullen raw score composite, 2) CARS score, 3) rate of total child communication, and 4) Parent Stress Index: Difficult Child Score. The independent variables used as maternal predictors were: 1) maternal IQ, 2) maternal education, 3) Beck Depression Total Score, and 4) PSI Parental Distress score. The Mullen raw composite was entered first in each regression model in order to control for child development. Six sets of regression models were evaluated in total, a maternal predictor model and a child predictor model for each of the three dependent variables of interest. We ran correlations for each of the variables as a first step; this was done to rule out any highly correlated variables which might not be uniquely contributing to our regression analyses. Tables 6 and 7 report the Pearson correlation coefficients between the maternal and child predictors and each of the dependent variables.

Most of the child variables (CARS total, Mullen raw composite, and rate of total child communication) were significantly correlated with the maternal responsivity composite, but

were not significantly correlated with behavior regulation or molar responsivity (see table 6). Of the maternal variables, maternal education and maternal IQ were the strongest predictors. Maternal education was significantly negatively correlated with behavior management and significantly positively correlated with molar responsivity. Maternal IQ was significantly correlated with both maternal responsivity and molar responsivity. Overall, the maternal predictors were most strongly correlated with the molar responsivity variable (see table 7). Examination of the correlations between these variables indicated that there were relationships among them, and that inclusion of the variables in the same regression model was appropriate.

Child Predictors

The child predictors were entered in separate blocks with the Mullen raw composite entered first to control for child development, followed by CARS score, PSI: Difficult Child score, and finally rate of child communication. Tests for multicollinearity and residuals were conducted to ensure that the data met the necessary regression assumptions and all values indicated no problem with the models. In terms of the maternal responsivity composite, the Mullen raw composite explained a significant amount of variance (35%). For the maternal responsivity composite outcome, the addition of the CARS and PSI: Difficult Child score to the model did not result in a significant *F* Change. The addition of rate of child communication did improve both models and significantly increased the percent of variance explained. Table 8 shows the final model statistics at each step when just the two significant variables are modeled. The adjusted *R* square of the final model for maternal responsivity composite was .69, indicating that 69% of the variance in maternal responsivity was accounted for by these two child variables. When both variables were in the model, only rate of child communication had a significant regression coefficient.

In terms of the molar responsivity variable, Mullen raw composite was not a significant predictor at the first step. The rate of total child communication predictor did significantly improve the model. With both predictors in the model, the *R* square was only .09 indicating that 9% of the variance in molar responsivity was explained. With both variables in the model, the regression weight for rate of child communication was significant. The two child predictors were not significant predictors of the behavior management variable.

In order to examine the impact of the full mutation within the mothers on the relationship between the child variables and maternal responsivity, a separate analysis was conducted with a reduced sample (dropping the three mothers with the full mutation and their children). See Table 9 for the summary for the reduced sample. The results were consistent with the results from the full sample with Mullen raw composite and rate of total child communication as the only significant predictors of maternal responsivity.

Maternal Predictors

The maternal predictors were entered in blocks with the Mullen raw composite first to control for child developmental level. This was followed by Beck Depression Total Score and the PSI: Parent Distress Score. Maternal education was entered next as a fourth block, followed by maternal IQ. Examination of the tests for collinearity in this full model

indicated that the Condition Index was greater than 30, suggesting that the high correlation between maternal education and maternal IQ was causing a multicollinearity problem. Since the correlations between maternal IQ and outcomes were stronger, we dropped maternal education from two of the models: maternal responsivity and molar responsivity. However, for the behavior regulation, we kept maternal education and dropped maternal IQ, since maternal education was the only significant correlate with behavior regulation. Table 10 represents the final regression models for each of the three maternal composites.

In terms of the maternal responsivity composite, Mullen raw composite was a significant predictor with an R^2 of .37. Adding Parental Distress to the model in step three increased the variance explained by 5% and was a marginally significant improvement. The addition of maternal IQ was a significant improvement and the overall model explained 56% of the variance in the maternal responsivity composite. In the final maternal responsivity model with four predictors, the parameter estimates for three of the four variables were significantly different from zero.

For the molar responsivity variable, 29% of the variance was explained by the maternal predictor variables. The strongest predictor was maternal IQ and it was the only variable that remained significant with the other variables in the model. After controlling for child developmental level, only the maternal education variable was a significant predictor of behavior management. In fact, the only model that significantly predicted behavior management was the model with maternal education in it (see table 10). With the maternal predictors in the model, 11% of the variance was accounted for, with maternal education as the only significant predictor.

We completed the analyses a second time excluding the mothers with the full mutation and their children. The results were similar to the full sample. In the reduced sample, Mullen raw composite accounted for a slightly higher proportion of variance in maternal responsivity composite scores and the addition of Beck Depression Total Score significantly improved the model. Maternal IQ was a significant predictor with the full mutation mothers removed, but its effect was not as pronounced. Table 11 provides information about the regression models for the three maternal composites with the reduced sample, including parameter estimates. Estimates were consistent across the samples.

Discussion

The purpose of this study was to determine the influence of maternal and child variables on the maternal responsivity of 55 mothers of young children with FXS. Specifically, we asked how maternal variables such as IQ (or education for the behavior management factor), stress and depression, and child variables such as developmental level and communication, affect maternal responsivity at both the molecular and molar level. The results indicated that two of the child variables significantly influenced molecular and molar responsivity: child developmental level and rate of child communication. In terms of the maternal variables, maternal IQ was the strongest predictor, correlating with both molecular and molar responsivity; however maternal education was the strongest predictor for behavior management.

Child Variables

Two of the child variables, child development level (Mullen raw composite) and child language level (rate of child communication) were significantly related to maternal responsivity. These two child variables accounted for 69% of the variance in the model. However, with both child variables in the model, only rate of child communication remained a significant and unique predictor of maternal responsivity.

The children in this study with higher rates of communication had more responsive parents; the more the children were communicating, the more opportunities the mothers had to respond. This does not imply a causal relationship, merely that the mothers likely had more opportunities to be responsive. One example of this type of relationship is the recode variable. In order for a mother to get credit for a recode, the behavior must follow a child communication act. Therefore, mothers of children with very low rates of communication would have fewer opportunities to expand on their child's communication. Nevertheless, communication level and molecular responsivity are not mutually exclusive. In this analysis, rate of child initiations did impact the amount of responsivity in terms of recodes, but not because the mothers were proportionally more responsive. Additionally, a child who has very low communication skills can still have a mother who uses frequent gestures and comments, and expands child communication attempts.

The presence of autistic symptoms as measured by the CARS did not impact molar or molecular responsivity. Certain child characteristics such as maladaptive behavior and unpredictable behaviors including impulsivity and aggressiveness have been shown to negatively influence maternal psychological well-being (Fidler, Hodapp, & Dykens, 2000; Hodapp, Fidler, & Smith, 1998); these behaviors are commonly associated with higher rates of autism. The presence of autism has been found to disrupt maternal-child interactions, and have an adverse effect on maternal responsivity (van IJzendoorn et al., 2007). Nevertheless, in this study, autistic symptoms did not appear to influence maternal responsivity after controlling for child developmental level and language ability.

Maternal responsivity does not function independently of the child's behavior and responsiveness. Either partner in the “dance” between parent and child is capable of disrupting the interaction and altering its very nature in ways that may extend out over a lifetime (Kelly & Barnard, 2000). Given that child communication and development are such robust predictors of responsivity in FXS, the need for early and intensive intervention is increasingly apparent. This study highlights the cyclical pattern of the mother-child relationship: early communication problems make it difficult for a mother to employ a highly responsive style of interaction, which impacts later language development. The majority of the children in this study were below chronological age expectations in terms of their language and cognitive abilities, so it is not the case that these findings are true for mothers of children with typical language development.

Maternal Variables

Maternal IQ, maternal education, current maternal depression (BDI-II), and the PSI Difficult Child score comprised the four maternal predictors in this analysis, after controlling for

child development. Maternal IQ was a significant predictor of maternal responsivity, accounting for 56% of the variance in molecular responsivity. The Beck Depression Total Score also contributed uniquely to the model; however, IQ had the strongest impact. In terms of molar responsivity, maternal IQ was also the strongest predictor of responsivity. The full model with all four variables explained 29% of the variance. However, IQ was the only variable that remained significant with the other variables included in the model.

For the behavior management factor, maternal education was the only significant predictor. However, the model with this variable only accounted for 11% of the variance in behavior management. The small amount of variance accounted for, and the fact that only one variable showed any association with this maternal behavior could have been due to the limitations of our data collection. That is, the behavioral management data were collected during the 25-minute direct observation window. During this short time period, we observed very little problematic child behavior that would warrant enough instances of behavior management responses to complete a meaningful analysis. The addition of challenging and frustrating measurement situations to our protocol for the child might have provided more variance and allowed a more meaningful analysis. Consequently, no conclusions should be drawn from this analysis due to these observational constraints.

As noted, maternal IQ and education were the strongest predictors of all three maternal factors. This is congruent with the findings reported in the larger literature on maternal responsivity (Bornstein & Tamis-LeMonda, 1989; Warren & Brady, 2007). We had a range of IQs for the mothers in our sample, and although the maternal education variable mean was 15 years for the full sample, several mothers did not complete high school. In short we had a relatively diverse sample in terms of education and IQ. This finding is potentially important given the variability associated with cognitive abilities in the broader fragile X phenotype. Women who are carriers of FXS have a greater likelihood of cognitive deficits in attention, verbal memory, and executive function (Freund et al., 1993; Sobesky et al., 1994), and women with the full mutation have an even greater likelihood of learning disabilities and low educational attainment.

Current maternal depression status and parenting stress measures did not uniquely impact molar responsivity although current depression did have a small impact on molecular responsivity. Several studies have reported a negative correlation between maternal depression and maternal responsivity (Goldsmith & Rothbart, 1996; Murray et al., 1996). The measure used in this analysis, the Beck Depression Total Score is a measure of current depression (Beck et al., 1996) within a 2-week period. Given that our analysis of maternal responsivity included a current examination of responsivity via live video recordings, we used a measure of depression linked to the mother's current state. Our limited findings probably reflect the constraints of our sample size and data collection methods. Although our sample was relatively large given the rare nature of FXS, in an absolute sense it was small enough to potentially constrain our analyses, thus not allowing for a valid analysis of depression and its impact on responsivity. Second our sample of maternal-child interactions was too limited to provide an opportunity to observe a relationship between depression and responsivity.

A strength of this study is the use of both molar and molecular coding systems in order to broadly examine responsivity in this sample of mothers. These two different measures of responsivity represent different aspects of a mother's parenting style. Nevertheless, the same child and maternal variables appear to influence both types of responsivity: maternal IQ, child developmental level, and rate of communication. However, the child variables accounted for a large percent (69%) of the variance for molecular responsivity, but only accounted for 9% of the variance for molar responsivity. In other words, in this study the child variables had more impact on how frequently a mother responded to their child, but not necessarily on how pleasant and warm her affect is toward her child.

Another strength is the relatively large sample size for a study of such a rare condition that is often not identified until children are age 3 or older. We were also able to recruit and retain 55 families from relatively diverse backgrounds (e.g., maternal education) given that it was a sample of convenience. However, a trade off to get this sample size was our inability to tightly control child age and potentially important variables such as race and ethnicity. While the sample size was relatively large for a study of FXS, it was not large enough to include additional variables such as history of intervention or gender due to limited power. These variables could be of importance in a study of responsivity.

This study has at least one important clinical implication. We found that child rate of communication had a significant impact on maternal responsivity, which suggests that increasing children's rate of communication should provide more opportunities for maternal responses. There are two ways to achieve this. One approach is to teach mothers who are proportionally unresponsive to increase the responsiveness to child initiations (Girolametto & Weitzman, 2006). Given the importance of early maternal responsivity, children with FXS or similar disorders whose parents do not naturally employ this style may be at added risk in terms of their social and communication development. There is empirical research indicating that highly responsive parenting styles can be acquired by parents of young children with disabilities through relatively modest amounts of training (Brady, Warren & Sterling, 2009). In addition to teaching parents to be more responsive, direct interventions that effectively increase the child's rate of initiations, provide more opportunities for parents to be responsive. Both intervention approaches can be combined in a single more comprehensive approach (Brady, Bredin-Oja, & Warren, 2008).

In summary, the results of this study indicate that both child and maternal variables influence maternal responsivity in biological mothers of young children with FXS. In terms of the molecular level of maternal responsivity, child variables such as language ability and developmental levels are the strongest influences; however, these same child variables are not as strongly related to responsivity at the molar level. In terms of the maternal factors examined in this study, maternal variables such as IQ are influential for both molecular and molar responsivity, while maternal education was a significant predictor of behavior management.

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Table 1

Means (and ranges) of Child Participants' Scores

Gender	N	CA of child in months	MSEL Composite Score	CARS total scores
Boys Mean Range	44	29.6 (11-40)	52.9 (49-93)	26.9 (16.5-36.5)
Girls Mean Range	11	21.6 (10-37)	71.1 (49-123)	22.3 (17-33.5)

2

Table Maternal Characteristics

	<i>N</i>	Education (years completed)	IQ	Beck Depression Inventory ¹	Parenting Stress Index ²
Total Sample Mean <i>SD</i>	55				
		15.30	106.73	6.69	79.27
		2.38	15.30	6.13	19.20
Premutation	52				
Mean		15.35	108.39	7.05	80.35
<i>SD</i>		2.41	13.33	6.11	18.57
Full Mutation	3				
Mean		14.33	82.33	.44	60.56
<i>SD</i>		2.08	24.68	.38	24.73

Note.

¹ Scores from the Beck Depression Inventory-II (BDI-II; Beck, Steer, & Brown, 1996) 5 women had scores indicating mild depression, 2 women had scores indicating moderate depression, and one woman had a score in the severe depression range. The mean was in the nonclinical range.

² Scores from the *Parenting Stress Index* (PSI; Abidin, 1995). The mean score was not in the clinical range of stress. However, 17 of the 55 women in this study had scores indicating clinically significant levels of stress.

3

Table Definition of Molecular Codes

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

4

Table Intraclass Correlation Coefficients (ICCs)

Variable	ICC
Child Variable:	
Child Rate of communication	.894
Maternal Responsivity:	
Gestures	.941
Request for Verbal Comply	.968
Comments	.946
Recodes	.799
<i>Average</i>	.914
Behavior Management:	
Request for Behavioral Comply	.925
Redirect	.777
Zap	.936
<i>Average</i>	.879

Table 5

Definition of Molar Codes

Behavior:	Definition:	Example:
Display of Positive Affect	Number of times a mom smiles directly at child	Smile must be directed at the child; mother's teeth must be visible
Warmth	Mom responds to child's verbal and nonverbal communication attempts; praise; enthusiasm	Mom 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	Mom who is highly responsiveness will be involved, but also patient for child's abilities.

6

Table Correlations between Child Predictors and Maternal Composites

	Mullen Raw Composite Score	Rate of Child Communication	PSI: Difficult Child Score	CARS Total Score
Maternal Responsivity	.589**	.828**	.145	-.396**
Behavior Management	.031	.173	.037	.001
Molar Responsivity	.063	.252	.198	.016

Note.

$N = 55$. * $p < .05$.

**
 $p < .01$.

Table 7

Correlations between Maternal Predictors and Maternal Composites

	Maternal IQ	Maternal Education	PSI: Parental Distress	BDI-II Total
Maternal Responsivity	.361 *	.124	.072	-.101
Behavior Management	-.178	-.319 *	.065	-.017
Molar Responsivity	.521 **	.310 *	.096	.042

Note. $N = 55$.*
p<.05.**
p<.01.

Table 8

Final Models with Two Significant Child Predictors: Full Sample

Predictor	Maternal Behaviors								
	Maternal Responsivity			Behavior Management			Molar Responsivity		
	<i>R</i> ²	<i>F</i>	β	<i>R</i> ²	<i>F</i>	β	<i>R</i> ²	<i>F</i>	β
Step 1	.347	26.63**		.001	.05		.004	.20	
Mullen ^a			.59**			.03			.06
Step 2	.338	52.70**		.048	2.46		.089	4.81*	
Mullen			-.02			-.20			-.25
Rate of Child Communication			.84**			.32			.43*
Total <i>R</i> ²	.686			.049			.093		
<i>n</i>	55			55			55		

Note.

*
p < .05.**
p < .01.^a Mullen variable was a composite score from the raw scores from the Mullen Scales of Early Learning.

Table 9

Final Models with Two Significant Child Predictors: Premutation Carriers only

Predictor	Maternal Responsivity			Maternal Behaviors Behavior Management			Molar Responsivity		
	R ²	F	β	R ²	F	β	R ²	F	β
Step 1 Mullen ^a	.394	30.50**	.63	.005	.22	.07	.005	.22	.068
Step 2	.312	48.77**		.024	1.14		.078	3.93*	
Mullen			.01			-.10			-.24
Rate of Child Communication			.83			.23			.42
Total R ²	.706			.029			.083		
<i>n</i>	52			52			52		

Note.

*
p<.05.**
p<.01.^a Mullen variable was a composite score from the raw scores from the Mullen Scales of Early Learning

Table 10

Final Models with Significant Maternal Predictors: Full Sample

Predictor	Maternal Behaviors								
	Maternal Responsivity			Behavior Management			Molar Responsivity		
	<i>R</i> ²	<i>F</i>	β	<i>R</i> ²	<i>F</i>	β	<i>R</i> ²	<i>F</i>	β
Step 1	.37	28.27**		.001	.05		.02	.76	
Mullen ^a			.61**			.03			.13
Step 2	.03	2.10		.000	.02		.00	.01	
Mullen			.63**			.03			.13
BDI			-.17			-.02			.002
Step 3	.05	4.04*		.011	.56		.01	.48	
Mullen			.63**			.03			.126
BDI			-.37*			-.12			-.09
PSI: Parent Distress			.30*			.15			.14
Step 4	.11	11.19**		.11	5.68*		.26	16.47**	
Mullen		2	.62*			-.03			.11
BDI			-.38**			-.18			-.11
PSI: Parent			.26			.13			.07
Distress Maternal IQ ^b			.34**			-.34*			.5288
Total <i>R</i> ²	.56			.12			.29		
<i>n</i>	55			55			55		

Note.

* $p < .05$.** $p < .01$.^a Mullen variable was a composite score from the raw scores from the Mullen Scales of Early Learning.^b Maternal education used for the behavior management in place of maternal IQ.

Table 11

Final Models with Significant Maternal Predictors: Premutation Carriers only

Predictor	Maternal Behaviors								
	Maternal Responsivity			Behavior Management			Molar Responsivity		
	<i>R</i> ²	<i>F</i>	β	<i>R</i> ²	<i>F</i>	β	<i>R</i> ²	<i>F</i>	β
Step 1	.42	32.74*		.005	.22		.02	.81	
Mullen ^a			.65**			.07			.13
Step 2	.06	5.05*		.002	.32		.01	.23	
Mullen			.69**			.08			.14
BDI			-.25*			-.05			-.07
Step 3	.05	4.84*		.009	.20		.02	1.05	
Mullen			.68**			.08			.14
BDI			-.47**			-.14			-.22
PSI: Parent Distress			.32*			.13			.21
Step 4	.04	3.71		.104	7.04*		.14	7.31**	
Mullen			.67**			.10			.12
BDI			-.44**			-.18			-.17
PSI: Parent			.28			.20			.13
Distress Maternal IQ			.20			-.33*			.38*
Total <i>R</i> ²	.57			.12			.19		
<i>n</i>	52			52			52		

Note.

*
p < .05.**
p < .01.^a Mullen variable was a composite score from the raw scores from the Mullen Scales of Early Learning.