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Maternal Responsivity Predicts Language Development in Young Children With Fragile X Syndrome

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Abstract

The relationship between early maternal responsivity and later child communication outcomes in young children with fragile X syndrome was investigated. Data were obtained from 55 mother–child dyads over a 36-month period. Performance data were obtained at each measurement point from video observations of four different contexts. These were coded for (a) child communication behaviors, (b) parent responsivity, and (c) behavior management behaviors. Results indicate that early maternal responsivity predicts the level of four important child language outcomes at 36 months of age after controlling for child developmental level and autism symptomology.

In this study we investigated the relationship between maternal responsivity and later communication and language development in a cohort of children with fragile X syndrome. *Maternal responsivity* refers to a healthy, growth-producing relationship characterized by warmth, nurturance, and stability as well as specific behaviors, such as contingent positive responses to child initiations. There is a substantial and growing body of evidence that cumulative exposure to a stable, highly responsive parenting style throughout the early childhood period is associated with a variety of child benefits in terms of language, cognitive, emotional, and social development (Landry, Smith, Miller-Loncar, & Swank, 1998; Landry, Smith, Swank, Assel, & Vellet, 2001; Siller & Sigman, 2008; Tamis-LeMonda, Bornstein, & Baumwell, 2001; Warren & Brady, 2007). Conversely, there is also substantial evidence that long-term exposure to a harsh and/or unresponsive and directive parenting style is associated with suboptimal outcomes across the same developmental domains (Bates, Pettit, Dodge, & Ridge, 1998; Hart & Risley, 1995). Parenting style itself appears to be affected by a number of variables, including parental emotional state (e.g., depression), beliefs and values, and maternal education level as well as variables such as the child's temperament and developmental level (Bornstein, 1995; Shapiro, Blacher, & Lopez, 1998).

Maternal Responsivity: Empirical Evidence

A substantial number of researchers have investigated the effects of maternal responsivity on child development. Viewed cumulatively, this body of research supports the contention that maternal responsivity plays an important role in enhancing child development (Landry, Smith, & Swank, 2006; Osofsky & Thompson, 2000). Children whose mothers display more responsive behavior during the first several years of life achieve language milestones earlier (Landry et al., 2001; Tamis-LeMonda et al., 2001), score significantly higher on cognitive tests (Landry, Garner, Swank, & Baldwin, 1996; Landry, Smith, Swank, & Miller-Loncar, 2000), develop better social skills (Calkins, Smith, Gill, & Johnson, 1998; Kochanska,

Forman, & Coy, 1999; Landry et al., 1998) and have fewer emotional and behavioral problems (Goldberg, Corter, Lojkasek, & Minde, 1991).

In a longitudinal study of 282 young children (103 full-term children, 102 low-risk preterm children, and 77 high-risk preterm children), Landry and her colleagues demonstrated that highly responsive parenting achieves its most substantial effects when it is sustained throughout the early childhood period (up to age 5 years). Children in this study who were exposed to highly responsive parenting early in development, but not later; or later in development, but not earlier, scored substantially lower on measures of language, cognitive, and social development compared with children who experienced ongoing, consistent responsiveness as well as maternal warmth over a period of several years (Landry et al., 2001). Thus, the impact of maternal responsiveness appears to grow cumulatively over many years, starting in infancy. Furthermore, the range of potential effects appears to be quite broad and includes language, social, emotional (i.e., attachment security), and cognitive development.

At the opposite end of the continuum from warm, highly responsive parenting is unresponsive and/or harsh parenting. Unresponsive parenting has been associated with low maternal education (Hooper, Burchinal, Erwick Roberts, Zeisel, & Neebe, 1998), depression (Rutter & Quinton, 1984), substance abuse (Osofsky & Thompson, 2000), and mild mental retardation (Miller, Heysek, Whitman, & Borkowski, 1996). Just as responsive parenting has been associated with accelerated growth in language, cognition, and social behavior, unresponsive parenting has been associated with lower growth trajectories (Tomasello & Farrar, 1986; Tomasello & Todd, 1983). Harsh parenting, which sometimes co-occurs with unresponsive parenting, has been shown to have a markedly negative impact on development and behavior (Dodge, Bates, & Pettit, 1990; Whitman, Borkowski, Keogh, & Weed, 2001).

The degree of parental sensitivity and responsiveness has also been shown to be predictive of outcomes in children with disabilities (Warren & Brady, 2007). In a large-scale longitudinal study of the development of children with intellectual disabilities and parent well-being, Hauser-Cram, Warfield, Shonkoff, and Krauss (2001) found that when mental age was controlled, the quality and frequency of mother–child interaction was the only significant correlate of communication skills at age 3 years. By 10 years of age, children whose parents' interaction scores were more positive had, on average, an advantage of approximately 10 months in communication skills. On the other hand, Wasserman and colleagues (Waserman, Allen, & Solomon, 1985) found that infants with disabilities whose mothers ignored them for a proportion of a free-play observation conducted when their child was 12 months of age had significantly lower intelligence scores at 24 months.

Yoder and Warren (1998, 2000, 2001) have also demonstrated that young children with intellectual disabilities who have highly responsive mothers achieve significantly greater gains in terms of later language development as a result of early prelinguistic communication intervention than do children with low responsive mothers. A number of other studies have demonstrated correlational relationships between maternal responsivity and style and the development of children with intellectual disabilities (Shapiro et al., 1998), including autism (Siller & Sigman, 2008). However, in general, the extent to which maternal responsivity and style observed across studies is an adaptive or maladaptive response to the behavior of the child with intellectual disabilities has been more the subject of speculation than research (Marfo, Dedrick, & Barbour, 1998; Osofsky & Thompson, 2000).

Maternal Responsivity: Theoretical Framework

The transactional model provides a theoretical framework for understanding how parenting style impacts cognitive and language development (Sameroff & Chandler, 1975; Sameroff & Fiese, 2000). This model presumes that early communication, social, and cognitive development is facilitated by bidirectional, reciprocal interactions between children and the environment. For example, a change in the young child, such as the onset of intentional communication around 9 months of age, may trigger a change in the social environment, such as increased linguistic mapping by their caregivers (linguistic mapping is known to facilitate receptive and productive language development: Gallaway & Richards, 1994; Warren & Walker, 2005). These changes then support further development in the child (e.g., increased vocabulary) and, subsequently, further changes by caregivers (e.g., more complex language interactions with the child). In this way both the child and the environment change over time and affect each other in reciprocal fashion as early achievements pave the way for subsequent development (Warren & Walker, 2005). Maternal responsiveness, operating through the reciprocally adjusting mechanism of the transactional model, may enhance early cognitive and language development by directly supporting the child's active exploration and engagement of the environment (Landry et al., 2000).

A transactional model may be well-suited for understanding early cognitive and language development because caregiver–child interaction can play such a unique role during this period (Warren & Brady, 2007). In the first few years of life, children's relatively restricted behavior repertoire allows changes in their behavior to be more salient and easily observable to caregivers (Warren & Yoder, 1998). This, in turn, may allow adults to be more specifically contingent with their responses to the child's developing interests and skills than is typically possible later in development, after the child's behavioral repertoire has become far more expansive and complex. Consider, for example, the relative ease with which a responsive parent can note instances of new learning in an infant or toddler (e.g., specifically acknowledging more words and word attempts) and how unlikely it is that the same parent can accurately account for most instances of new learning just 2 years later (Sokolov & Snow, 1994).

The true power of the transactional model is its cumulative impact across lengthy periods of time. Consider the relentless manner by which cumulative advantages and deficits in experience can develop across the first few years of life. For example, an input difference in positive affect expressed by a parent toward their child of 10 events per day (a difference of less than 1 event per waking hour, on average) would result in a cumulative difference of 10,950 such events over a 3-year period. If a child who experiences less positive affect also experiences cumulatively more negative affect (e.g., "Stop that," "Get out of there," Shut your mouth up," "You're a bad boy"), it becomes relatively easy to conceive of combinations of these qualitative and quantitative experiential differences contributing to deficits in exploratory behavior, self-concept, language, and social development (Warren & Walker, 2005). Furthermore, we have substantial evidence that such large cumulative deficits occur in typically developing young children and that these differences strongly correlate with important indicators of development later in childhood (e.g., vocabulary size, IQ, reading ability, and school achievement: Feagans & Farran, 1982; Gottfried, 1984; Hart & Risley, 1992; Walker, Greenwood, Hart, & Carta, 1994).

Influence of Child and Parent Characteristics

In part because they often display low rates of initiation and responsiveness themselves (Hauser-Cram et al., 2001; Yoder, Davies, & Bishop, 1994), young children with developmental delays, such as those associated with fragile X syndrome, are perhaps more likely to experience relative deficits in various types of environmental input compared to

typically developing children, despite the best intentions of their caregivers. For example, in a study with mother–Down syndrome infant dyads, Slonims and McConachie (2006) found that at 8 weeks of age, these infants were already significantly less communicative and lively relative to typically developing infants. At this very early point in development, mothers of these children were indistinguishable from mothers of typically developing children. By 20 weeks of age, however, mothers were significantly less sensitive and more remote than were the mothers of typically developing children. Thus, these children were already on different paths in terms of their experiential histories of cumulative responsivity. Is a similar process already at work with children with fragile X syndrome? It is not possible to do a similar study with these children at the present time (or children with autism) because diagnosis typically occurs when children are at least 2 years of age. We do know that the presence of autism typically disrupts mother–child interaction in general and maternal responsivity specifically (van IJzendoorn et al., 2007). Because somewhere between 25% and 40% of boys with fragile X syndrome also have autism, it may be that at least some level of disruption occurs early in their development.

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). Initiating and maintaining a highly responsive interaction style with a child who has fragile X syndrome, autism, or any of a number of other disorders can be highly challenging, even for a parent with the best of intentions (Stormont, 2001). A number of phenotypic characteristics of fragile X syndrome may be disruptive to parental responsivity alone or in combination with other characteristics. These include gaze avoidance or atypical eye gaze, hypersensitivity to sensory input, social anxiety and shyness, perseveration and repetitiousness, stereotypical behavior, unintelligible speech, and problems with conversational discourse (Abbeduto, Brady, & Kover, 2007; Bailey, Hatton, & Skinner, 1998; Sterling & Warren, 2008). In addition, a substantial number of children with fragile X syndrome also fall on the autistic 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.

In addition to the challenges to highly responsive parenting that the child may bring to the mother–child relationship, recent research on the neuropsychological and emotional functioning of women who are carriers of fragile X syndrome suggests that although they generally have intelligence scores in the typical range, they often have subtle cognitive deficits in executive function, attention, visual spatial skills, and verbal memory (Freund, Reiss, & Abrams, 1993; Mazzocco, Pennington, & Hagerman, 1993; Reiss, Freund, Baumgardner, Abrams, & Denckla, 1995; Sobesky, Hull, & Hagerman, 1994). Women who are premutation carriers of fragile X syndrome tend to display more social anxiety and are more affectively labile (Sobesky et al., 1994). Women who have fragile X syndrome have been shown to have significantly higher levels of lifetime depression than do women with similar developmental delays (Thompson, Rogerness, McClure, Clayton, & Johnson, 1996). These risk factors for mothers of children with fragile X syndrome are also well-established risk factors for lowered maternal responsiveness (Osofsky & Thompson, 2000). For example, maternal mental health has been repeatedly shown to negatively influence the way mothers interact with their young children (Goldsmith & Rothbart, 1996; Murray, Fiori, Hooper, & Cooper, 1996). Stress and anxiety have also been shown to affect maternal responsivity (Osofsky & Thompson, 2000).

Phenotypes evolve and develop over time through the process of genotype–environment interaction. At any given point in time, an *individual phenotype* is the collective manifestation of anatomical, neurological, and behavioral traits resulting from the past history of interaction and ongoing interaction of heredity and environment of that phenotype. In the case of child development, heritable characteristics of the child may evoke a stable pattern of particular parental responses that have a cumulative impact on the child's behavior and development over lengthy periods of time (Abbeduto, Evans, & Dolan, 2001; Collins, Maccoby, Steinberg, Hetherington, & Bornstein, 2000; Sameroff & Fiese, 2000). In the case of fragile X syndrome, it may be that the high heritability of problematic behavior, including impaired social and cognitive engagement, could suppress parental responsiveness and over time evoke a stable pattern of parent directiveness and low responsivity (Warren, 2004). In a sense, the child's phenotype creates its own learning environment, one that is far from optimal. As Landry and her colleagues have shown with high-risk preterm children (Landry et al., 2000; Smith, Landry, & Swank, 2000, 2000) and Hart and Risley (1992, 1995) with typically developing young children, it is the stable, cumulative nature of parental responsivity over many years that bestows the greatest development advantage.

Our main focus in this study was to determine the relationship between maternal responsivity and communication and language development over time in a sample of young children with fragile X syndrome. We were particularly interested in discerning whether predictive relationships exist between maternal responsivity at approximately 24 months and later measures of child communication and language development, controlling for child cognitive level and autism symptomology. The specific research question was: Does early maternal responsivity predict level and growth at 36 months in both proximal and distal measures of communication and language development when differences in developmental level and autism symptomology are controlled?

Method

Subjects

Fifty-five children with fragile X syndrome between 11 and 48 months of age at the first observation and their biological mothers were recruited from across the United States. Measures were collected at three assessment points. Children were between 26 and 64 months at the second data point and between 40 and 76 months at the third. Recruitment efforts included advertising at national conventions, use of a national research registry housed at the University of North Carolina-Chapel Hill, networking with fragile X syndrome family support groups, and advertising via a fragile X syndrome parent list serve.

This was a sample of convenience because fragile X syndrome is a rare disorder with few symptoms at birth, and children are often not identified until age 3 or older. Although desirable, we were not able to tightly control either age of entry into the study or potentially important socioeconomic status (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 full-mutation fragile X syndrome. Table 1 provides descriptive information on the child participants. Composite scores from the Mullen Scales of Early Learning—hereafter called the Mullen Scales (Mullen, 1995) provide an estimated developmental age in months. The Childhood Autism Rating Scale—CARS (Schopler, Reichler, & Renner, 1988) provides a well-validated measure of autistic symptoms (Rellini, Tortolani, Trillo, Carbone, & Montecchi, 2004). If a child scores in the range of 30 to 36 on the CARS, he or she is considered to be in the mild to moderate range for autism. A score above 36 is considered an indicator of severe autism. However, we did not use the score on

the CARS as a diagnostic indicator, but simply as a measure of the presence of autistic-like behavior during the time points when we collected data for each participant.

Three of the mothers also had full mutation fragile X syndrome, and all the other mothers were carriers. We included the 3 full mutation mothers in the study because they represent a small but important segment of mothers with children who have fragile X, thus making our sample more representative. Each mother completed the Wechsler Adult Intelligence Scale —third edition (Wechsler, 1997). The mean IQ for the full sample of mothers was 107; and the range of scores, 55 to 130, was substantial. The 3 mothers with full mutation fragile X syndrome had IQs of 55, 89, and 103. 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 was married, and 88% were Caucasian. They ranged in education from 6 parents with a high school education or less to 53% who had graduated from college. The full mutation mother 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.

Measures

Data were collected on the child with fragile X syndrome and his or her biological mother. Direct measures of maternal responsivity and child communication observed during mother–child interactions in four different contexts were included.

Performance measures of child communication and maternal responsivity— We videotaped four different interactional contexts and later coded them to obtain the performance measures. Five min of each of the following contexts were videotaped in the dyads' home: reading a book together, making and eating a snack together, unstructured play with toys of their choosing, and a 30-min sample in a naturalistic context. We judged that this range of contexts and the length of time samples in each context was sufficient to give us a representative measure of the child's language and the mother's behavior toward the child. For the naturalistic context, we instructed parents to conduct an everyday activity, such as putting dishes away, folding clothes, or playing together. The three 5-min contexts and two 5-min segments from the 30-min naturalistic context were digitized for coding, yielding a total of 25 min of interaction. This amount of interaction coded is similar to that reported in other studies of maternal responsivity (Warren & Brady, 2007).

The digitized video files were then coded using Noldus™ Observer software, version 5.1 (Noldus, 2002). Two child measures coded were rate of total communication and rate of number of different words observed during the interaction. We obtained the first child measure 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. The rate of number of different words consisted of the number of different words or signs during an observation divided by the total time.

Each videotaped observation file was also coded for maternal responsivity using the Noldus™ software. We coded the mother's speech 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 behaviors 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—Paid graduate students were trained to identify and code the behaviors listed above 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 this, we compared transcripts, 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, namely, maternal responsivity, maternal behavior management, and two proximal child communication outcomes, we calculated intraclass correlation coefficients (ICCs), using the absolute agreement and single measure values 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 (see Table 3). We used this procedure to determine whether the consensus coding procedure biased the data.

The ICCs were high for child rate of communication behaviors, .98 calculated between primary and reliability and .98 between primary and consensus coded data. They 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.

Other child measures: The Mullen Scales—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 an *SD* of 15. The subscale scores reported within the analyses are based on raw scores, whereas the reported composite score is the standardized score.

Childhood Autism Rating Scale—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*) to 4 (*severely abnormal for age or developmental level*). The total score can be used as a continuum of autistic behavior. Also, performance on the measure can be interpreted as nonautistic (total score of 15 to 29.5), mildly or moderately autistic (total score of 30 to 36.5), and severely autistic (total score of 37 or higher) The examine*r*s administered the CARS at each of three time points as a measure of autistic-like behavior.

Results

Preliminary Data Analysis

Principal components analysis—Our emphasis in the current paper is on the influence of early maternal behavior on subsequent child language outcomes. Preliminary analysis revealed a substantial level of correlation among the seven maternal codes for young children ages 19 to 37 months. Therefore, we conducted a principal components analysis (Gorsuch, 2003), which reduced the seven maternal codes to two principal components for observations made over this age range. The first component included gesture use, requests for verbal complies, comments, and recodes (verbal interpretations of the child's communication act). We 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"). We labeled this *component behavior management*. In order to combine scores that were quite varied in scale into a single component score, we

computed z scores for each indicator and then averaged the z scores across the scores within the factor for each child. Asymptotically, these composite scores should have a mean of zero for both composites and an SD of .25 for the maternal responsivity composite, where we are combining four scores with pairwise correlations of approximately .50. These averaged z scores for maternal responsivity and behavior management were used in subsequent analyses.

Hierarchical linear models (HLM)—The majority of the children had limited communication at the initial observation as indicated by low scores on all four communication variables. Figures 1 through 4 show the observed data for Mullen Receptive Language raw score, Mullen Expressive Language raw score, rate of number of different words, and rate of total communication for each child over the three observations, respectively. Most children increased in their communicative behaviors over the course of the study, with more rapid growth occurring at younger ages and a leveling off of the growth as children aged. Thus, models with linear and quadratic growth were initially examined. Figures 1 through 4 also indicate that there were some girls who scored consistently at the upper end of the distribution. This potential gender difference should have been addressed in the modeling procedures. However, there were not enough females ($n = 11$) participating in the study to evaluate separate models for males and females (gender effect), so all models were run twice: once for the total sample and then again for boys only. An inspection of the changes in the model parameters with the two samples provided some indication of the effect the girls had on the models.

As noted in the *Subjects* section above, children participating in the study ranged in age from 11 to 48 months at the first observation and were assessed at approximately 18-month intervals thereafter. The average time between Observations 1 and 2 was 17.5 months and between Observations 2 and 3, 15.35 months. The average time between Observations 2 and 3 was less because some of the older children were assessed at shorter intervals to prevent them from aging out of some of the measures. This large relative range in age at the first observation period was included in the models by using age in months as the time metric. All observations regardless of age were included in the growth models. Because the majority of the children were observed by 36 months (only 7 children had their first observation at an older age), age was centered on 36 months by subtracting 36 from the child's age at each observation so that all intercepts are interpreted as level of communication at 36 months.

Modeling Approach

Children obviously experience significant growth in communication over time, and this growth may have quadratic trends. Figures 1 through 4 indicate that linear and quadratic growth was indeed observed in the repeated measures over time in the current sample. The three observations for each individual in the current data can be viewed as repeated measurements (Level 1) within individuals (Level 2). Thus, a two-level model with repeated measures at the lowest level and persons at the highest level was most appropriate, and this type of model analysis was well-suited to display the varying ages at observation reflected in these data. A set of models was evaluated for each child-language outcome using SAS PROC MIXED. Fixed effects terms for intercept, linear growth, and quadratic growth were included in a model with random effects for intercepts and slopes (linear growth). That is, the trajectory of growth with age was modeled in the fixed effects while allowing individual intercepts and slopes to vary (random effects). Probability levels for each of the effects are shown in Tables 4 through 7; however, the decisions concerning which effects to include in the models were made by comparing models using two types of indices: the deviance statistic, or change in the −2 log likelihood, and the Bayesian information criterion. Deviance statistics are distributed asymptotically as chi square with degrees of freedom

equal to the difference in the number of parameters estimated by the two models. Smaller Bayesian information criterion values indicate better fit. Thus, models were chosen based on the results of the chi-square deviance tests and the size of the Bayesian information criterion values. Once the growth model was established, we added additional child control variables (described below) to the models to yield base models.

It is well-established in the empirical literature that child characteristics such as developmental level and the presence of autistic symptoms can influence later language development. Because the focus of our analyses was the effect of maternal responsivity on later communication development, we wanted to control for important child differences. Thus, we employed the child's Mullen Scales Composite score as a control variable in the models for the rate of number of different words and rate of total communication. The Mullen Scales Composite was not used as a control variable in the models of Receptive and Expressive Language because these subscales are part of the Mullen Scales Composite. An examination of the Mullen Composite score over time revealed that many children who first were assessed before the age of 24 months had much higher Mullen Composite scores at their first assessment compared with their later Mullen Composite scores. We hypothesized that the limited number of items applicable to young children might be resulting in an inflated Mullen Composite score at this first assessment. Therefore, the score obtained at the first observation for each child at 24 months or older was used as a control variable in the models.

CARS scores were obtained for the children at each observation to assess autistic characteristics. For about one third of the children, CARS scores fluctuated somewhat over the three observations. Some increased; others bounced around from lower to higher and back down. Because of the fluctuation, the average of the last two CARS scores was computed to yield the most representative score. Because CARS scores greater than or equal to 30 indicate the presence of autism symptomology (Schopler et al., 1988), we created a dichotomous variable from the CARS scores reflecting this situation: Scores less than 30 were coded as 0, indicating the absence of autism symptomology; the others were coded as 1, indicating the presence of autism symptomology. We refer to this latter group of children as having autism symptomology or as having a positive autism symptomology score. In this study, 18 children had average CARS scores over their last two observations at or above 30. This dichotomous predictor was also entered into the growth model to control for behavior associated with autism that is not captured in overall child development (Mullen Scales Composite).

The influence of both the Mullen Scales Composite and the autism symptomology scores on intercept, linear growth, and quadratic growth was evaluated simultaneously for each outcome. Comparisons between the models were made using the deviance statistic described above. Nonsignificant terms were dropped from the models until all the parameters left resulted in significantly improved models. This model became the base, or comparison, model to which the effects of maternal variables (responsivity and behavior management) on intercept, slope, and quadratic growth were added. Nonsignificant maternal effects were dropped from the model until only terms that significantly improved the fit of the model remained. The base, or comparison, model and the final model with maternal responsivity as a predictor for each child language outcome are presented in Tables 4 through 7. The modeling processes enabled us to determine whether maternal responsivity and behavior management significantly influenced language outcomes after controlling for differences in development and autism symptomatology. To keep the timing of the assessments consistent, we used the data from the first observation at age 24 months or older to compute a maternal responsivity score for each mother.

Behavior Management

The addition of behavior management to the base models for the child language outcomes did not significantly improve any of the models. Therefore, all subsequent discussion of the effects of maternal variables involves only maternal responsivity as a predictor.

Receptive Language Models

The raw score from the Mullen Scales Receptive Language subscale was the outcome variable used to model growth in receptive language. The only control variable in these analyses was the autism symptomology indicator.

Receptive language models for total sample (boys and girls)—The best-fitting growth model for the Mullen Scales Receptive Language subscale (Table 4, Model 1a) included significant fixed effects for intercept, linear growth, and quadratic growth. Receptive language scores at 36 months were significantly lower, χ^2 (1, $N = 55$) = 30.4, $p < .0001$, for children with autism symptomology (average = 16.22) than for other children with fragile X syndrome (average = 22.57). Similarly, rate of linear growth was significantly different between groups, χ^2 (1, $N = 55$) = 23.1, $p < .0001$. Children with autism symptomology increased their receptive language scores by .31 every month on average, while other children with fragile X syndrome increased by .56 every month on average. For both groups, the estimate for the quadratic term was −.003, indicating that the rate of growth in receptive language was slowing over time. Significant random effects for intercept and slopes were also included in the base model. Adding the effect of maternal responsivity on the intercept to the base models for the total sample resulted in a significantly improved model, χ^2 (1, $N = 55$) = 5.6, $p < .05$. In the total sample, for every unit change in maternal responsivity, the receptive language score at 36 months was increased by 1.51. These effects can be illustrated graphically by looking at the expected outcomes for children with differing values on the predictor of interest using the parameter estimates obtained from the model. For example in Figure 5, the effects of CARS score (0 or 1) and maternal responsiveness (±1 *SD* from the mean) on receptive language trajectory are depicted. The solid dark line represents the predicted receptive language trajectory for a hypothetical child with a low CARS score and a mother scoring 1 *SD* above the mean on maternal responsivity. The intercept value of 36 months is noted with the vertical line.

Receptive language model for boys only—The boys-only best-fitting base model was very similar to the total sample in that intercept, linear, and quadratic growth with random intercepts and slopes was the best-fitting growth model. The children with CARS scores greater than 30 had significantly lower intercepts, χ^2(1, $N = 44$) = 26.0, $p < .0001$, and significantly slower rates of linear change, χ^2(1, $N = 44$) = 17.2, $p < .0001$, just as in the total sample base model. Adding the effect of maternal responsivity to the boys-only model for receptive language did not improve the model significantly, χ^2(1, $N = 1$) = 2.7, $p = .10$ For boys, for every unit change in maternal responsivity, the Receptive Language score was increased by 1.17.

The difference in the parameter estimates for the total sample (1.51) and boys only (1.17) suggests that in the current sample the relationship between maternal responsivity and receptive language may be stronger for girls than boys (although sample size limitations did not allow for an analysis of interaction effects). Figure 5 illustrates the effect of maternal responsivity on Receptive Language scores for the total sample when the effect of autism symptomology was included in the model. Children whose mothers engaged in higher levels of responsive behavior scored higher in receptive language.

Expressive Language Models

Raw scores from the Mullen Scales Expressive Language subscale were used to model growth in expressive language. Autism symptomology status was used as a control variable.

Expressive language models for total sample (boys and girls)—The best-fitting growth model for Mullen Scales Expressive Language raw scores (Table 5, Model 1a) included significant fixed effects for intercept, linear growth, and quadratic growth. Expressive language scores at 36 months were significantly lower for children with autism symptomology, χ^2 (1, $N = 55$) = 28.3, $p < .0001$, and linear rate of growth was significantly slower for children with autism symptomology, $\chi^2(1, N = 55) = 36.0$, $p < .0001$, than for other children with fragile X syndrome. For both groups, the estimate for the quadratic term was $-.003$, indicating that the rate of growth in expressive language was slowing over time. Significant random effects for intercepts and slopes were also included in the base model. Adding the effect of maternal responsivity on the intercept, slope, and quadratic growth to the base models for the total sample resulted in a significantly improved model, $\chi^2(3, N = 55) = 15.1$, $p < .01$. In the total sample, for every unit change in maternal responsivity, the expressive language score at 36 months was increased by 2.88, $p < .01$. The positive parameter estimate for the effect of maternal responsivity on linear growth indicates that as children aged, the effect of responsivity was somewhat increased. The negative parameter estimate for the effect of maternal responsivity on the quadratic indicates that as children aged, the impact of maternal responsivity lessened. Adding maternal responsivity to the base model for expressive language reduced the variance in intercepts by 17%, from 24.03 to 19.90. The residual variance was also reduced by 5%.

Expressive language model for boys only—As in the total sample, the best-fitting growth model for Mullen Expressive Language for boys included fixed effects for intercept, linear growth, and quadratic growth. Expressive language scores at 36 months and linear rate of growth were significantly lower for children with positive autism symptomology scores than for other children with fragile X syndrome, $\chi^2(1, N = 44) = 28.6$, $p < .0001$, and $\chi^2(1, N = 44) = 23.2$, $p < .0001$, respectively). For both groups, the estimate for the quadratic term was $-.003$, indicating that the rate of growth in expressive language was slowing over time. Significant random effects for intercepts and slopes were also included in the base model. Adding the effect of maternal responsivity on the intercept, slope, and quadratic growth to the base models for the boys-only model resulted in a significantly improved model, $\chi^2(3, N = 44) = 13.8$, $p < .01$. For every unit change in maternal responsivity, the expressive language score at 36 months was increased by 2.78, $p < .01$, the rate of linear growth was increased by .126, $p < .05$, and the term for quadratic growth was decreased by .005, $p < .05$. Adding maternal responsivity to the base model for expressive language reduced the variance in intercepts by 21%, from 14.77 to 11.63. The residual variance was also reduced by 7% with the addition of maternal responsivity to the model.

For both boys and the total sample, children whose mothers were more responsive had increased levels of expressive language at 36 months compared with children whose mothers were less responsive. Figure 6 shows the predicted expressive language score for mothers with high and low levels of responsivity (one *SD* above the mean and one *SD* below the mean). The effect of maternal responsivity was most pronounced for children in the lower range of the age distribution.

Rate of Number of Different Words

Model for rate of number of different words for the total sample—The best-fitting base model for the rate of number of different words in the total sample (Table 6, Model 1a) included a fixed effect for linear growth. Development (i.e., the Mullen Scales Composite

score) significantly influenced the rate of number of different words at 36 months, $\chi^2(1, N = 55) = 47.3, p < .0001$. The addition of the effect of autism symptomology scores on the intercept in the model indicated that children with low autism symptomology scores were using more different words than were children with autism symptomology, $\chi^2(1, N = 55) = 10.8, p = .001$. The linear rate of change (slope) in the number of different words was significantly different between the two groups, $\chi^2(1, N = 55) = 12.7, p < .001$, with children scoring low on the CARS having larger slopes. The growth model also included significant random effects for intercepts and slopes. For the total sample, maternal responsivity significantly influenced rate of number of different words at 36 months, $\chi^2(1, N = 55) = 17.9, p < .0001$. The addition of maternal responsivity to the model decreased variance in intercepts by 35%.

Model for rate of number of different words for boys only—The best-fitting growth model for boys only included a fixed effect intercept at 36 months and a fixed effect for linear growth. Development and autism symptomology status significantly impacted rate of number of different words at 36 months (intercept), $\chi^2(1, N = 44) = 36.5, p < .0001$, and $\chi^2(1, N = 44) = 9.6, p < .01$, respectively, and autism symptomology status significantly influenced linear rate of change (slope) $\chi^2(1, N = 44) = 14.1, p < .001$. As in the growth model for the total sample, random effects for intercepts and slopes were included. In the boys-only model, adding the influence of initial maternal responsivity on the level of the rate of number of different words at 36 months, significantly improved the model beyond the base model, $\chi^2(1, N = 44) = 6.5, p = .01$. The addition of maternal responsivity to the growth model for boys reduced the residual variance by 16%.

Both models indicate that after including child factors in the model (Mullen Scales and autism symptomology), the effect of maternal responsivity was significant. Children whose mothers were more responsive had increased rates of number of different words. Figure 7 illustrates the predicted rate of number of different words.

Models for Rate of Total Communication

This model looks at growth in all forms of child communication, including prelinguistic gestures, vocalizations, and words.

Model for rate of total communication in the total sample—The best-fitting base model for rate of total communication in the total sample (Table 7, Model 1a) had a fixed effect for linear growth growth. Mullen Scales Composite scores (development) significantly influenced number of words at 36 months, $\chi^2(1, N = 55) = 23.4, p < .0001$. Presence of autism symptomology had a significant effect on intercept, $\chi^2(1, N = 55) = 18.9, p < .0001$, and linear change (slope), $\chi^2(1, N = 55) = 4.8, p < .05$. Children without autism symptomology had a higher rate of communication and a larger slope. The base model also included significant random effects for intercept and slope. For the total sample, maternal responsivity significantly influenced both the rate of communication at 36 months, $\chi^2(1, N = 55) = 5.9, p < .05$, and the linear growth, $\chi^2(1, N = 55) = 11.7, p < .001$.

Model for rate of total communication for boys only—The best-fitting base model for rate of total communication in boys had the same terms as the model for the total sample. Maternal responsivity did not, however, significantly influence rate of total communication for the boys only. Figure 8 illustrates the predicted rate of total communication for children whose mothers were high or low (± 1 SD) on maternal responsivity.

Three of the mothers in the sample themselves had full-mutation fragile X syndrome. Table 8 provides individual scores for these three mothers and their child. The child scores for the

CARS and Mullen Composite and the four outcome variables are presented as are the mother's IQ, education level, and relative ranking in terms of the maternal responsivity component. Total sample means are also provided for each variable as a reference point.

Discussion

In this study we investigated the relationship between maternal responsivity and later communication and language development in a cohort of children with fragile X syndrome. Results indicate that early maternal responsivity predicted the level of two important proximal child language outcomes (rate of total communication and rate of number of different word) at 36 months of age, controlling for child developmental level and autism symptomology, and on distal measures of receptive and expressive language development, controlling only for autism symptomology. Our results support both transactional theory and empirical studies suggesting that exposure to relatively high levels of maternal responsivity can significantly impact the communication and language development of all children, including those with genetically based disorders such as fragile X syndrome.

It has been well-established in the literature that maternal responsivity does not function independently of the child's behavior and responsiveness. Either member of the dyad can disrupt interaction. Initiating and maintaining a highly responsive parenting style with a child who has fragile X syndrome and/or autism can be difficult for any parent, regardless of their intentions (Girolametto, Weitzman, & Clements-Baartman, 1998). Certain phenotypic characteristics of children with fragile X syndrome alone or in combination with other disorders (e.g., social anxiety, hypersensitivity to sensory input, level of developmental delay) may make this challenge especially difficult. Nevertheless, this study shows that even when we controlled for development and/or autism symptomology, the children of mothers who engaged in higher levels of responsivity as we defined it benefited significantly from early exposure to this style as measured by their level of communication and language development at 36 months.

The general pattern of effects was similar across the four outcome measures, as is evident in Tables 4 through 7. In each case, adding maternal responsivity to the model that already controlled for developmental level and/or autism symptomology resulted in a significantly improved model of level at 36 months. Children whose mothers engaged in higher levels of maternal responsivity tended to score higher than children of mothers who scored lower in maternal responsivity. This relationship was stronger with the total sample than with the boys-only sample, but the overall effects followed this pattern irrespective of gender. Although these data indicate that the presence of autism symptomology clearly inhibits receptive language growth, high maternal responsivity did appear to lessen the impact to some extent as shown in Figure 5.

A similar effect is evident for expressive language (Figure 6). Although the presence of autism symptomology clearly impacts development, high maternal responsivity lessens the impact to some extent. In terms of both the proximal measures of communication (rate of total communication and rate of number of different words), and distal measures (Receptive and Expressive Language subscales of the Mullen Scale), children with low autism symptomolgy and low maternal responsivity grew at a substantially accelerated pace compared to children with high autism symptomology scores and either high or low maternal responsivity. That is, although high maternal responsivity offsets the impact of autism somewhat, there was no evidence that it offset it completely (see Figures 5 through 8). Furthermore, the impact of autism was so strong that for the most part, even children with low responsivity and low autism outperformed children with high parental responsivity

and autism symptomology. In other words, the impact of having autism and fragile X syndrome appears greater than having fragile X syndrome and a low responsive mother.

We did not have a sufficient number of girls to do a meaningful analysis of their data separate from the boys. Therefore, we created a kind of ad-hoc control for gender by analyzing the data for the total sample and for boys only. It is well-documented that fragile X syndrome generally impacts girls less than boys. Figures 1 through 4 provide a visual picture of the extent of overlap between the girls and the boys on the four outcome measures. In general, these figures show some overlap but also indicate that girls tend to perform at higher levels on these measures than do boys. The results of our boys-only and full-sample HLM models for each variable suggest that the general patterns were similar for the two groups overall but that there were consistent differences in intercept and slope, exactly as one might expect.

Although the pattern of results across the four measures is similar, there are important differences in the nature of the two distal and two proximal measures. The Mullen Scales Receptive and Expressive Language measures index language knowledge that has accumulated over time. Thus, scores on these subscales may be viewed as reflecting the effects of the child experiencing a prolonged relative difference in maternal responsivity over months or years. The proximal measures (rate of total communication and rate of number of different words) index performance in the four measurement contexts and likely reflect accumulated knowledge to some extent and the relative interaction between the child and the parent. That is, these measures are more susceptible to context effects. That the pattern of results obtained is relatively consistent across these two types of measures suggests that they are also relatively robust.

How did the children of the full mutation mothers do on the proximal and distal measures? The answer to this question depends on the measure and the child. The children of the two lowest IQ full mutation mothers were females and, thus, were likely buffered to some extent from the full direct biological impact of fragile X syndrome because of their gender. In fact, the female whose mother had the lowest maternal IQ in the study (IQ = 55) had scores above the group average on two of the four language outcome variables. However, she also had a Mullen score that was more than 20 points below the mean for females in the group. Furthermore, her mother was the lowest ranked in the entire sample in terms of maternal responsivity. The other female's mother was also very low on responsivity, and her pattern of scores across all measures except the CARS was low. The son of the highest IQ full mutation mother had scores above the average or near average on all measures, and his mother's responsivity score was slightly above average relative to the full sample. Overall, the 3 mothers' responsivity levels appeared to track their IQ level. All 3 children had low CARS scores (22, 23, and 25).

In sum, because the sample of full mutation mothers was very small, no a priori conclusions should be drawn about their parenting skills or the potential of their children on measures of language development and use. Nevertheless, the maternal responsivity levels of the 2 low IQ mothers and their daughter's generally low overall performances (relative to the sample) suggests that mothers' full-mutation status may be associated with additional risk for full-mutation children. On the other hand, the relatively strong performance by the son of the full-mutation mother with an average IQ underscores the degree of variability associated with full-mutation status in females. Therefore, research with a much larger sample of full-mutation mothers is necessary to make any meaningful conclusions about the impact of this status on maternal parenting skills or child outcome.

Although we conducted the same analyses on the behavior management component as we did on the maternal responsivity component, we found no significant relationships between this component and any of the four language outcome measures. All mothers, of course, used the behaviors that made up this component. However, the frequencies of use were generally much lower than the maternal responsivity behaviors. Although this finding did not surprise us, we believe it is inappropriate to draw any conclusion from it.

This study has certain inherent strengths and limitations. In terms of strength, the mere fact that we were able to recruit 55 families with young children who have fragile X syndrome and retain them in a longitudinal study is a clear strength as is the relative diversity of the sample along several dimensions (e.g., SES, maternal education level). However, to recruit a sample of this size, it was necessary for us to take a flexible approach in terms of age of entry. This resulted in a large degree of variability on this dimension. We have managed this variability by using actual age in the analyses and then centering the data at 36 months. This approach facilitates interpretation of the parameters and the effects of early responsivity on these parameters, which, in turn, facilitates interpretation of the effects of maternal responsivity on later development.

Another strength of the study is that we were able to control for both development (using overall Mullen Scales scores) and autism symptomology (using a dichotomous CARS-score variable). Both the Mullen Scales and the CARS have a high degree of validity and reliability. However, the CARS is not generally viewed as sufficient to diagnose a child as having autism. Thus, we have been careful to employ it as a measure of autism symptomology. Finally, by employing principal component's analysis in combination with HLM, we have been able to quantitatively estimate the effects of maternal responsivity on later development as opposed to limiting the analysis to a correlational approach or to a less robust analysis of each of the separate outcome measures.

We believe that this study has at least one important clinical implication. Given the importance of early maternal responsivity, children with fragile X syndrome or similar disorders whose parents do not naturally employ this style may be at added risk in terms of their social and communication development. However, there is a body of empirical research indicating that highly responsive parenting styles can be acquired by parents of young children with disabilities through relatively modest amounts of training (Warren & Brady, 2007). For example, Girolametto et al. (1998) reported success in establishing highly responsive parenting styles with parents of young children with Down syndrome based on as little of 20 hours of parent training over a few months. Furthermore, there is virtually no risk associated with these interventions. This study and others (McCauley & Fey, 2006) suggest the potential value of providing this type of training to all parents of young children with developmental delays early in their child's life.

In conclusion, the results of this study demonstrate a predictive relationship between early maternal responsivity on both distal and proximal measures of communication and language use in children with fragile X syndrome when controlling for autism symptoms and development. This serves as a demonstration of how a specific, but potentially ubiquitous environmental variable, parenting style, can influence the development of important skills in children with a genetic disorder associated with lifelong intellectual and developmental disabilities. Further, results suggest that efforts should begin early in the life of children with this and similar disorders to assure that parents and significant others engage in this style of parenting on a daily basis.

Acknowledgments

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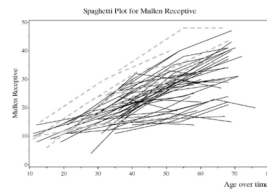
Figure 1.
Observed Mullen Receptive Language raw scores for girls (dashed lines) and boys (solid lines).
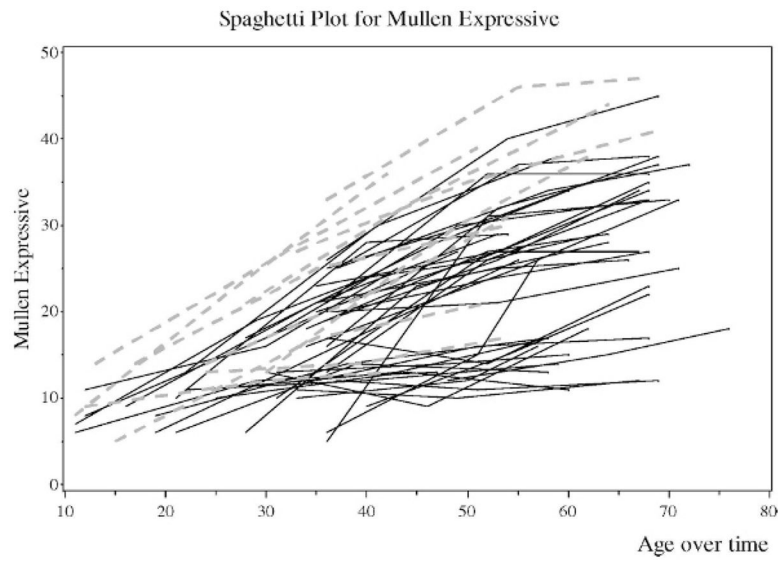
Figure 2.
Observed Mullen Scales of Early Learning Expressive Language raw scores for girls
(dashed lines) and boys (solid lines).
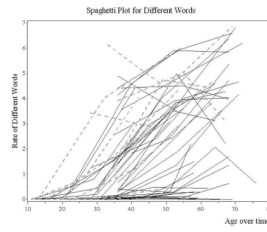
Figure 3.
Observed rate of number of different words for girls (dashed lines) and boys (solid lines).
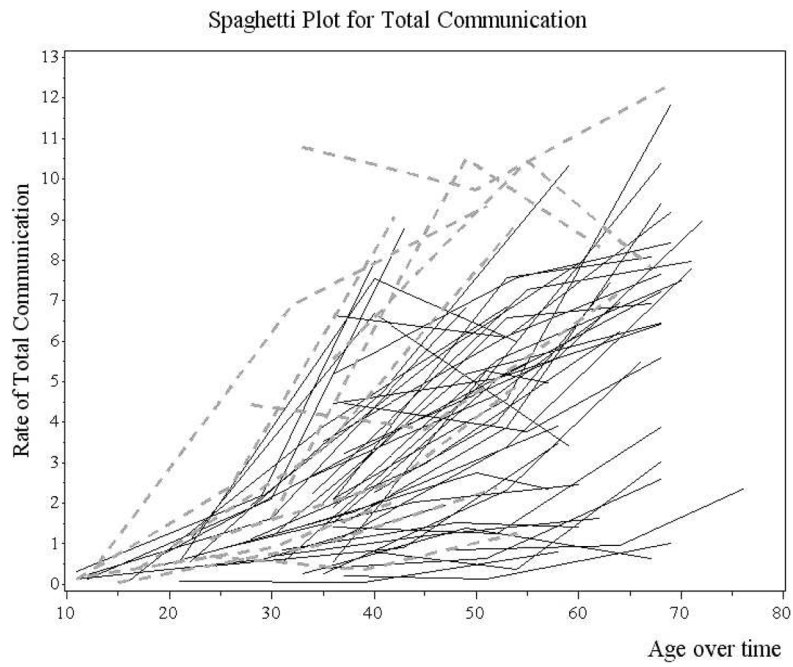
Figure 4.
Observed rate of total communication for girls (dashed lines) and boys (solid lines).

Figure 5.
Predicted receptive language for children with high and low responsive mothers. Solid black line = low CARS, high responsivity; black dashed line = low CARS, low responsivity; solid gray line = high CARS, high responsivity; dashed gray line = high CARS, low responsivity.
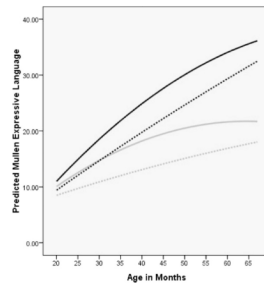
Figure 6.
Predicted expressive language for children with high and low responsive mothers. Solid black line = low CARS, high responsivity; black dashed line = low CARS, low responsivity; solid gray line = high CARS, high responsivity; dashed gray line = high CARS, low responsivity.
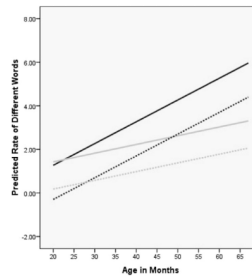
Figure 7.
Predicted rate of number of different words for children with high and low responsive mothers. Solid black line = low CARS, high responsivity; black dashed line = low CARS, low responsivity; solid gray line = high CARS, high responsivity; dashed gray line = high CARS, low responsivity.
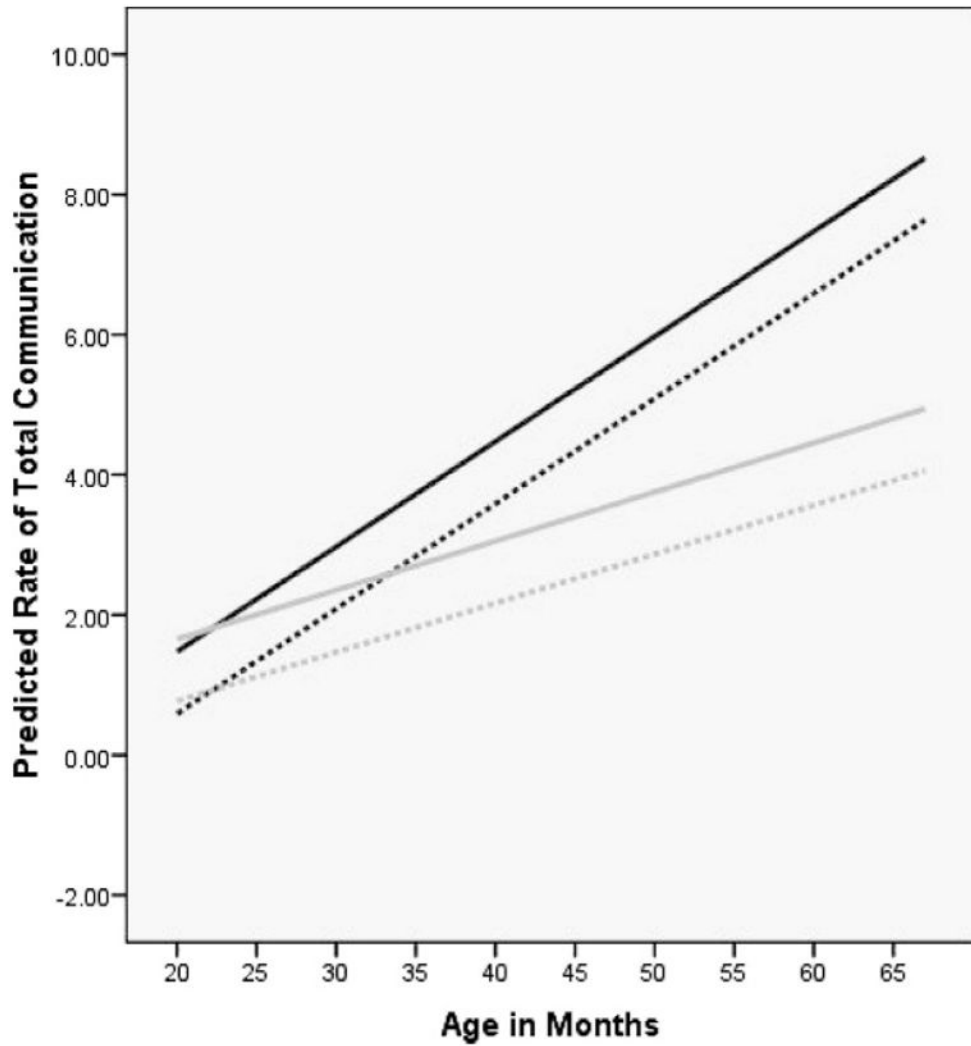
Figure 8.
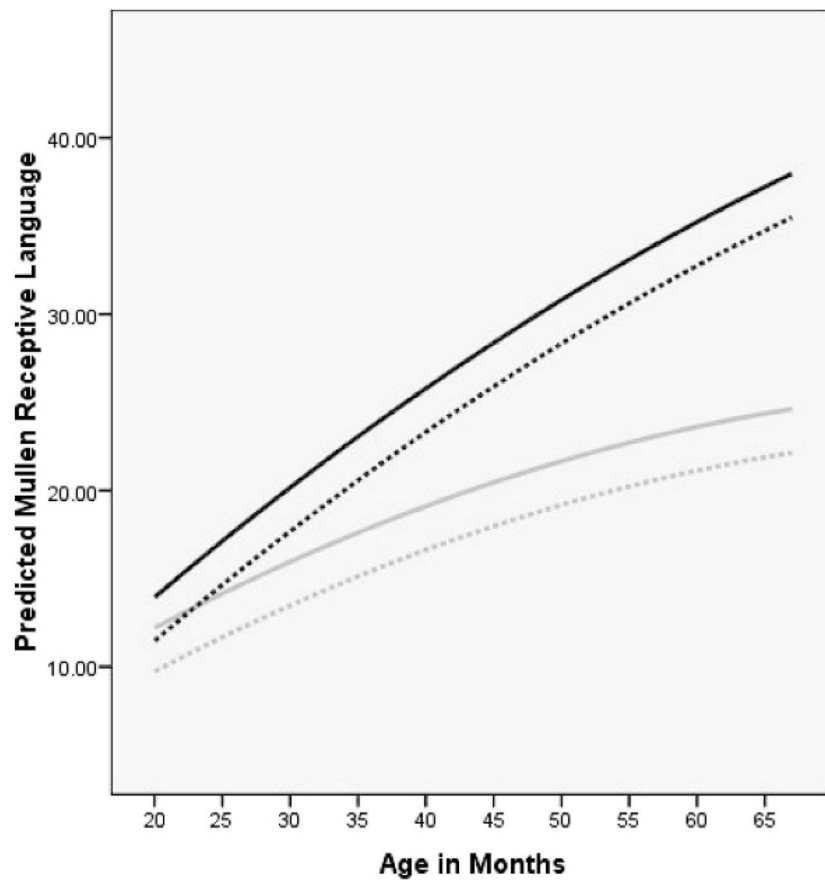Predicted rate of total communication for children with high and low responsive mothers. Solid black line = low CARS, high responsivity; black dashed line = low CARS, low responsivity; solid gray line = high CARS, high responsivity; dashed gray line = high CARS, low responsivity.

Table 1

Means of Child Participants' Scores at First Observation

Child measure	Boys (*n* = 44)		Girls (*n* = 11)	
	Mean	Range	Mean	Range
CA of child at first observation[a]	29.6	11–40	21.6	10–37
MSEL[b] Composite score	52.9	49–93	71.1	49–123
CARS[c] total scores	26.9	16.5–36.5	22.3	17–33.5

[a]In months.

[b]Mullen Scales of Early Learning.

[c]Childhood Autism Rating Scale.

Table 2

Definition of Maternal Codes

Behavior	Definition	Example
Gestures	Sign language, gestures (e.g., "come here," "stop," "no"), tapping, clapping, or knocking	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?"
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."

Table 3

Intraclass Correlation Coefficients Across the Three Observations

	Observation		
Variable	**1**	**2**	**3**
Child rate of communication	.89	.83	.96
Rate of no. of different			
words	.99	.99	.99
Average	.94	.91	.98
Gestures	.94	.97	.99
Request for verbal comply	.97	.98	.99
Comments	.95	.98	.98
Recodes	.80	.99	.98
Average	.91	.98	.99
Request for behavioral comply	.93	.95	.98
Redirect	.78	.81	.86
Zap	.94	.92	.96
Average	.88	.90	.90

Table 4

Parameter Estimates (Est.) for Receptive Language for Total Sample and Boys Only

| | Total sample | | | | Boys only | | | |
| | Model 1a: Base model | | Model 2a: Base model with maternal responsivity | | Model 1b: Base model | | Model 2b: Base model with maternal responsivity | |
Variable	Est.	SE[a]	Est.	SE	Est.	SE	Est.	SE
Fixed effects								
Growth variables								
Intercept	16.22	.94***	16.46	.91***	15.97	.88***	16.05	.87***
Linear growth	.31	.04***	.31	.04***	.30	.05***	.30	.05***
Quadratic growth	−.00	.00**	−.00	.00**	−.00	.00*	−.00	.00*
Control variables								
Autism on intercept	6.35	1.14***	5.69	1.13***	5.38	1.08***	5.01	1.09***
Autism on linear	.25	.05***	.25	.05***	.25	.06***	.24	.06***
Predictor variables								
Maternal responsivity on intercept			1.51	.64*			1.17	.74
Maternal responsivity on linear								
Random effects								
Intercept	11.32	3.05***	10.15	2.85***	6.57	2.46**	6.41	2.39**
Linear growth	.02	.01**	.02	.01**	.02	.01**	.02	.01**
Residual	8.07	1.46***	8.13	1.48***	8.27	1.65***	8.23	1.65***
Model fit								
−2 log likelihood	936.2		930.6		737.4		734.7	
BIC[b]	968.1		966.7		767.6		768.8	

[a]Standard error.

[b]Bayesian information criterion.

*
$p < .05$.

**
$p < .01$.

***
$p < .001$.

Table 5

Parameter Estimates (Est.) for Expressive Language for Total Sample and Boys Only

| | Total sample | | | | Boys only | | | |
| | Model 1a. Base model | | Model 2a. Base model with maternal responsivity | | Model 1b. Base model | | Model 2b. Base model with maternal responsivity | |
Variable	Est.	SE[a]	Est.	SE	Est.	SE	Est.	SE
Fixed effects								
Growth variables								
Intercept	13.62	1.26***	14.16	1.17***	12.80	1.17***	13.31	1.07***
Linear growth	.27	.05***	.266	.05***	.27	.05***	.26	.06***
Quadratic growth	-.00	.00**	-.00	.00**	-.00	.00*	-.00	.00*
Control variables								
Autism on intercept	7.02	1.54***	5.52	1.47***	6.48	1.44***	5.02	1.36***
Autism on linear	.28	.05***	.29	.06***	.25	.06***	.27	.07***
Predictor variables								
Maternal responsivity on intercept			2.88	.85**			2.78	.94**
Maternal responsivity on linear			.07	.04			.13	.06*
Maternal responsivity on quadratic			-.00	.00*			-.01	.00*
Random effects								
Intercept	24.03	5.54***	19.90	4.73***	14.77	4.37***	11.63	3.64***
Linear growth	.02	.01**	.020	.01**	.03	.01*	.02	.01**
Residual	8.04	1.56***	7.63	1.48***	9.55	1.98***	8.84	1.87***
Model fit								
−2 log likelihood	976.4		961.3		772.4		758.6	
BIC[b]	1008.4		1005.4		802.4		800.2	

[a] Standard error.

[b] Bayesian information criterion.

* p < .05.

**
$p < .01.$

***
$p < .001.$

Table 6

Parameter Estimates (Est.) for Rate of Number of Different Words for Total Sample and for Boys Only

| | Total sample | | | | Boys only | | | |
| | Model 1a. Base model | | Model 2a. Base model with maternal responsivity | | Model 1b. Base model | | Model 2b. Base model with maternal responsivity | |
Variable	Est.	SE[a]	Est.	SE	Est.	SE	Est.	SE
Fixed effects								
Growth variables								
intercept	.99	.27***	1.01	.23***	1.29	.27***	1.16	.27***
Linear growth	.04	.01*	.04	.01*	.03	.01*	.03	.01*
Control variables								
Mullen on intercept	.10	.01***	.08	.01***	.17	.03***	.14	.03***
Autism on intercept	.95	.33**	.80	.29**	.77	.30*	.73	.28*
Autism on linear	.07	.02***	.06	.02**	.07	.02***	.06	.02***
Predictor variables								
Maternal responsivity on intercept			.76	.17***			.50	.20*
Random effects								
Intercept	.72	.23**	.47	.17**	.38	.16**	.32	.14*
Linear growth	.00	.00***	.00	.00***	.00	.001**	.002	.001***
Residual	.87	.16***	.82	.14***	.63	.13***	.60	.12***
Model fit								
−2 log likelihood	564.3		546.4		401.5		395.0	
BIC[a]	596.3		582.5		431.8		429.0	

[a]Standard error.

[b]Bayesian information criterion.

*
p < .05.

**
p < .01.

***
p < .001.

Table 7

Parameter Estimates (Est.) for Rate of Total Communication for Total Sample and Boys Only

| | Total sample | | | | Boys only | | | |
| | Model 1a. Base Model | | Model 2a. Base model with maternal responsivity | | Model 1b. Base model | | Model 2b. Base model with maternal responsivity | |
Variable	Est.	SE[a]	Est.	SE	Est.	SE	Est.	SE
Fixed effects								
Growth variables								
Intercept	1.91	.35***	1.93	.34***	2.14	.40***	2.11	.41***
Linear growth	.07	.02***	.07	.02***	.06	.02***	.06	.02***
Control variables								
Mullen on intercept	.10	.02***	.08	.02***	.15	.04***	.14	.05**
Autism on intercept	1.21	.44**	1.10	.43*	1.06	.44*	1.06	.44*
Autism on linear	.09	.02***	.08	.02***	.09	.02***	.09	.02***
Predictor variables								
Maternal responsivity on intercept			.54	.25*			.10	.31
Random effects								
Intercept	1.08	.39**	.96	.37**	.75	.33*	.78	.34*
Linear growth	.00	.00*	.00	.00*	.00	.00*	.00	.00*
Residual	1.93	.34***	1.89	.33***	1.53	.30***	1.53	.30***
Model fit								
−2 log likelihood	652.9		648.1		493.2		493.1	
BIC[b]	685.0		684.1		523.5		527.1	

[a]Standard error.

[b]Bayesian information criterion.

*
  $p < .05$.

**
  $p < .01$.

***
  $p < .001$.

Table 8

Measures of Three Full-Mutation Mothers and Their Children

Measure	Female	Female	Male	Total sample means (gender)
CARS[a] score	22	23.5	25	26.9 (M) 22.3 (F)
Mullen Composite score–Time 3	50	50	49	52.9 (M) 71.1 (F)
Rate of different words–Time 3	.71	4.29	4.71	3.64
Rate of total communication–Time 3	2.38	4.90	9.38	5.98
Expressive Language raw score	21	31	27	27.67
Receptive Language raw score	30	27	29	30.59
Maternal IQ	89	55	103	107
Maternal education	15	12	16	15
Maternal responsivity rank (percentile)	9th	2nd	55th	

[a]Childhood Autism Rating Scale.