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# The effect of sustained maternal responsivity on later vocabulary development in children with Fragile X Syndrome

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## Abstract

**Purpose—**The research question addressed was whether sustained maternal responsivity (a parent-child interaction style characterized by warmth, nurturance and stability as well as specific behaviors such as contingent positive responses to child initiations) was a significant variable predicting vocabulary development of children with FXS through age 9 years.

**Method—**Fifty-five mother-child dyads were followed longitudinally when children were between 2 and 10 years of age. Measures of maternal responsivity and child vocabulary were obtained at regular intervals starting at age 2.9 years. Sustained responsivity was indicated by the average responsivity measured over observations 2–5. Responsivity at the first time period, autism symptoms, and cognitive development were used as control variables.

**Results—**After controlling for development and autism symptoms, we found significant effects for sustained responsivity on receptive vocabulary, expressive vocabulary, and the rate of different words children produced through age 9.

**Conclusions—**Maternal responsivity, which is typically a variable of interest during early childhood, continues to be a significant variable, predicting vocabulary development through the middle childhood period. Thus, responsivity is a potential target for language interventions through this age period.

### Keywords

fragile X syndrome; language development; maternal responsivity; longitudinal

Children with fragile X syndrome (FXS) face challenges in terms of communication and language development in the middle childhood period. Most boys and many girls with FXS show significant lags in all areas of language and communication development with substantial variability among children of similar ages who all have the diagnosis of FXS (Abbeduto, Brady, & Kover, 2007; Hessl et al., 2008; Roberts, Mankowski, et al., 2008). In a previous study with the same cohort of children with FXS reported on here, we found that early maternal responsivity was significantly related to early language development (Warren, Brady, Sterling, Fleming, & Marquis, 2010). That is, children who had more

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responsive mothers had better language skills compared to children who had less responsive mothers. At the time of the Warren et al. study, the children were between 2 and 5 years of age. The purpose of the current study is to report on effects of sustained maternal responsivity on child vocabulary development and use for this same cohort through age 9.

## Language Development in Fragile X Syndrome

FXS is an inherited disorder caused by the mutation of the Fragile X mental retardation 1 (*FMR1*) gene and is commonly associated with intellectual disabilities and language impairments. Children with FXS typically have language profiles that show deficits relative to children of comparable chronological age (Abbeduto et al., 2007; Brady, Skinner, Roberts, & Hennon, 2006; Roberts, Mankowski, et al., 2008). Children with FXS have difficulties in all areas of language including comprehension, production, and pragmatic skills (Finestack & Abbeduto; 2010, Price, Roberts, Vandergrift, & Martin, 2007). While there is a fair amount of variability in language skills in FXS, production is a significant problem, particularly for boys with FXS. Several studies have reported that boys are below mental age expectations on language production measures, particularly measures derived from conversation language samples (Finestack, Sterling, & Abbeduto, 2013; Roberts, Mirrett, & Burchinal, 2001). Some variability is associated with gender. Girls are more mildly affected because they have two X chromosomes and normally only one chromosome is affected by the fragile X mutation. The remaining (unaffected) chromosome provides some protective function in terms of gene expression. However, since boys only have one X chromosome, the consequences of having a mutation on the *FMR1* gene is more substantial in terms of cognitive and language development (Abbeduto et al., 2007).

Variability is not only attributable to sex differences, as large variations in expressive language have also been found in boys with FXS. For example, Finestack & Abbeduto (2010) found ranges of 51 to 151 C-units (a measurement unit composed of an independent clause with all modifying clauses) in a group of high functioning boys with FXS. Similarly, Roberts, et al., (2007) reported standard deviations between 15 and 22.9 on mean scores for a standardized test of expressive vocabulary for boys with FXS between the ages of 3 and 14 years. Determining the sources of this variability is important for understanding and treating language disorders in children with FXS.

The presence and degree of autism symptoms also appears to be related to language development in FXS. Numerous studies have documented lower language levels for children who have co-morbid autism in addition to FXS, in comparison to individuals with FXS but not autism (e.g., Bailey, Hatton, & Skinner, 1998; Philofsky, Hepburn, Hayes, Hagerman, & Rogers, 2004). In our earlier study, we also found that children with more autism symptoms had lower language outcomes (Warren et al., 2010). However, Kover and Abbeduto (2010) found that the amount of talk and linguistic diversity (number of different words in a 50 C-unit sample) were delayed in all children with FXS, and comparable between children with and without co-morbid autism. Kover and Abbeduto's results were based on language sample analyses.

The few longitudinal studies completed to date indicate that variability in language abilities persists throughout childhood and is observable across both receptive and productive language. Roberts, et al. (2001) reported that variability continued over the course of a longitudinal study completed between the ages of 20 and 86 months. A major finding in this study was that the amount of delay increased over time for both receptive and expressive language, but more so for expressive language.

In the current investigation we focused on receptive and expressive vocabulary development, as opposed to more comprehensive language measures, for several reasons. First, other studies have found that vocabulary is generally commensurate with other facets of language development (Abbeduto et al., 2003; Roberts, Chapman, Martin, & Moskowitz, 2008). Second, vocabulary may be a particularly good index for overall language when children are still largely at the one-word stage (MLU less than 1) because children at these beginning stages of expressive language may show floor effects on other aspects of language development such as morphology and syntax. (Several children in the current investigation met this description.) Third, it is difficult to find a single comprehensive language measure that is appropriate across the wide range of ages (i.e. 2–9 years) and abilities of the children in the current study. Fourth, vocabulary, as measured by the rate of number of different words, was an outcome measure in the earlier study (Warren et al., 2010), as well as in other research studies on the effects of maternal responsivity (Girolametto, Weitzman, Wiigs, & Pearce, 1999); hence we are able to compare current findings to previous findings. Finally, vocabulary size is widely considered to be strongly associated with environmental inputs (e.g., parenting, reading) and thus is an ideal measure to study the effects of maternal responsivity.

## Maternal Responsivity and FXS

Maternal responsivity is defined as a “healthy, growth-producing relationship characterized by warmth, nurturance, and stability as well as specific behaviors, such as contingent positive responses to child initiations” (Warren et al., 2010; Warren & Brady, 2007). A highly responsive parent or caregiver provides a facilitative context for promoting many aspects of child development, including communication (Landry, Smith, & Swank, 2006). This study focused on the contribution of verbal responsivity to later child language outcomes. Increased contingent responding to child communication attempts, including imitation and expansion, is associated with better language outcomes for children (Baumwell, Tamis-LeMonda, & Bornstein, 1997; Bornstein, Tamis-LaMonda, Hahn, & Haynes, 2008; Girolametto et al., 1999; Masur, Flynn, & Eichorst, 2005; Yoder & Warren, 1998, 1999). Specifically, the type of behaviors - described as “follow-in” behaviors - that maintain a child’s focus of attention have been linked to positive vocabulary outcomes in children (McCathren, Yoder, & Warren, 1995; McDuffie & Yoder, 2010; Tomasello & Farrar, 1986; Watson, 1998). For example, if a child is playing with a rattle and the child’s mother comments, “oh you have a rattle,” the mother’s comment could be described as following-in with the child’s interest.

In contrast, low levels of maternal responsivity and/or excessive negative and redirecting types of responses have been found to have negative relationships with children’s language growth. Children who have mothers who provide relatively low amounts of verbal input have lower vocabulary growth over time (Hart & Risley, 1995; Huttenlocher, Haight, Bryk, Seltzer, & Lyons, 1991). Relatively high amounts of directive input that does not follow-in on the child's focus of attention has also been negatively correlated to language growth (Mahoney & Neville-Smith, 1996). Other aspects of a nonresponsive style, such as use of prohibitions and negative commands, have also been linked to poor language outcomes in children (Taylor, Donavan, Miles, & Leavitt, 2009). For example, frequent commands to do something outside of the child's immediate focus of attention may be counterproductive for language growth.

Much of the responsivity literature has examined how maternal responsivity during early childhood affects child language outcomes during this period (Masur et al., 2005; Pan, Rowe, Singer, & Snow, 2005; Pungello, Iruka, Dotterer, Mills-Koonce, & Reznick, 2009) while few studies have considered long term effects of maternal responsivity. A study by Siller and Sigman (2008), however, followed a group of children with autism between the ages of 3 and 8 years. Language gain over this period was measured with one of three standardized language tests. Results showed that children whose parents were more responsive during initial play interactions had significantly higher gain scores than children of parents who were less responsive (Note the authors use a different term, *synchronicity*, but the construct is similar to what we refer to as responsivity).

Although a great deal of emphasis has been placed on the importance of early maternal responsivity, consistent responsivity throughout childhood may provide the optimal interactive environment for promoting continued growth in language and other developmental domains. Continuous adjustment of the types and amounts of responsivity provided in response to changing child behaviors is the hallmark of healthy, adaptive parenting (Hauser-Cram, Warfield, Shonkoff, & Krauss, 2001; Legerstee, Varghese, & van Beek, 2002). Landry and colleagues (Landry, Smith, Swank, Assel, & Vellet, 2001) examined patterns of responsivity during infancy and early childhood and then examined the relationship between these patterns and growth in child cognitive and social skills. Children who experienced consistently high maternal responsivity throughout their first 5 years had better outcomes than children whose mothers were inconsistent or consistently low. However, it may be difficult for a mother to remain responsive over time if her child does not produce clear communication signals for long periods and/or develops at a very slow pace. Thus, it is particularly important to examine early responsivity as well as sustained responsivity over time for children with intellectual and developmental disabilities and in particular, children with communication impairments.

The current study investigated the effects of verbal maternal responsivity on child language development between the ages of 2 and 9 years in a group of children with FXS. Previously we reported that maternal responsivity was significantly related to receptive and expressive communication levels for this cohort when the children were between the ages of 2 and 5 (Warren et al., 2010). That is, children who had more responsive mothers early in development had significantly higher language scores and higher vocabulary growth during mother-child interactions. These effects were most pronounced for children with fewer autism symptoms. We extended our longitudinal study through middle childhood (ages 8–

10) in order to investigate the effect of sustained maternal responsivity for children's vocabulary development. Our hypothesis was that sustained maternal responsivity will significantly contribute to child vocabulary attainments through the middle childhood period, controlling for differences in initial cognitive development, autism symptoms, and early maternal responsivity.

## Methods

The methods employed in this study were approved by the Institutional Review Board of the University of Kansas.

### Participants

The participants of this study were a cohort of 55 children with FXS and their mothers. Each dyad has been part of our longitudinal study for 7 years. Participants represented a sample of convenience and were recruited from across the United States through advertisements at national conventions, use of a national research registry, and networking with the community of families who have a child with FXS.

**Maternal characteristics—**The mean age of mothers was 33.5 years at the first observation, ranging between 20.5 and 41.75 years. At the final observation the mean age was 38.8 years with a range of 25.9 to 47.1 years.

Three of the mothers had the full mutation fragile X syndrome, and 52 are premutation carriers. The *FMR1* gene is made up of trinucleotide (CGG) repeats, and elevated repeats beyond 55 signify either the premutation (55–200 repeats) or the full mutation (> 200 repeats). In the case of the full mutation, the elevated repeat sequence reduces or entirely shuts down the production of FMRP, a protein which is believed to play an important role in typical brain development and functioning (Hagerman, 2002; Rogers, Wehner, & Hagerman, 2001). In most individuals with the premutation, FMRP levels are within normal limits.

At the beginning of the study, each mother completed the Wechsler Adult Intelligence Scale – Third edition (Wechsler, 1997). The mother's IQs ranged widely from 55 to 130, with the mean score being 107. The IQs of the three mothers with full mutation fragile X syndrome were 55, 89, and 103. At the time of the final data point reported in this study, maternal education ranged from 9 to 20 years, with the average being 15.5 years, and 61% having graduated from college. Seventy-two percent were married. The majority of the mothers were Caucasian (91%), with small percentages of mothers identifying themselves as African American, Pacific Islander, or bi-racial. Household income showed considerable variability, with 11% reporting incomes of $30,000 or lower, and 33% reporting annual income of greater than $100,000. Most of the mothers worked outside the home, either part-time or full-time (61%).

**Child characteristics—**Forty-four boys and 11 girls participated in the study. The age of children at recruitment and first observation varied between 11 months and 4 years. In order to compare child and maternal data across similar ages, the current analyses include all data points beginning when children were 2 years old or older (median age of first data point was 2.9 years). All data collected for each child between the ages of 2 and 10 years were included in the growth analysis.

Nearly all of the children had some degree of cognitive impairment, as expected with FXS. Scores from the Brief IQ subtest of the Leiter-R International Performance Scale-Revised (Roid & Miller, 1997) are presented in Table 1. These scores were obtained at the time we collected Peabody Picture Vocabulary Test (PPVT-4;Dunn & Dunn, 2007) and Expressive Vocabulary Test (EVT-2; Williams, 2007) data, approximately age 8. Although data from another measure of cognitive development (the Mullen Scales of Early Learning, Mullen, 1995) was available at earlier time points, the nonverbal portions of this test are limited to visual processing and do not reflect the breadth of nonverbal skills comprised by the Leiter-R. Therefore, we relied on the Leiter-R scores to measure nonverbal cognitive status in this study.

Children in this study also had varying degrees of autism symptoms, as reflected in their scores on the Childhood Autism Rating Scale (CARS;Schopler, Reichler, & Rochen Renner, 1988), also presented in Table 1. Both the Leiter-R and the CARS are described below. The CARS scores were used to measure autism symptoms at the time of each home visit in our analyses, and not for diagnostic purposes. However, 18 participants did have an official diagnosis of autism in addition to FXS; 11 were diagnosed by a pediatrician, 1 by a developmental pediatric neurologist, and 6 by a psychologist. Parents indicated that 5 children were diagnosed based on the CARS and 5 were diagnosed based on the Autism Diagnostic Observation Schedule (ADOS;Lord, Rutter, DiLavore, & Risi, 1999). Parents could not recall the instrument used to diagnose the remaining 8 children.

## Measures

**Dependent variables—**The focus of this study was vocabulary development, and three different measures of vocabulary were included: the PPVT-4 the EVT, and the number of different words recorded during videotaped mother-child interactions (NDW). The PPVT-4 is a 228-item standardized measure for individuals between 2:6 and 90+ years of age that assesses an individual’s single word receptive vocabulary skills. Individuals are asked to identify a picture by pointing to or stating a letter from a field of four. The EVT-2 is a 190-item standardized measure for individuals between 2:6 and 90+ years of age. Individuals are asked to give a single word to label a picture or provide a synonym for the target word. We used raw scores from both of these measures in order to reduce floor effects associated with standard scores. Although age equivalent (AE) scores provide descriptive information, they are not appropriate for statistical analyses because they are ordinal (as opposed to interval), thus, age equivalent scores are not of an equal interval. For example, if Child 1 has an AE score of 12 months and Child 2 has an AE score of 24 months, it cannot be assumed that Child 2 has skills that are 12 months more advanced than Child 1 (Maloney & Larrivee, 2007; Mervis & Klein-Tasman, 2004).

Vocabulary produced during mother-child interactions was obtained from recordings across four different contexts: joint book reading, making and eating a snack together, unstructured play, and a daily living activity such as grooming or household activity. Five minutes from the first three listed contexts and 10 minutes from the daily living context were combined to yield a total of 25 minutes of interaction. We used 10 minutes from the daily living context because we observed twice as long in that context. These contexts were selected to provide a variety of activities with opportunities for different vocabulary production, yet maintain contextual measurement consistency across dyads. Digitized video files were coded and transcribed using Noldus Observer XT software (Noldus Information Technology, 2002).

Each child utterance was transcribed, and codes were used to indicate if words were signed as opposed to spoken. However, very few signs were observed. Transcripts were then analyzed with SALT, the Systematic Analysis of Language Transcripts (Miller & Chapman, 1985), to determine the number of different words produced by each child at each observation. The mean time of observations was 24.8 minutes and the standard deviation was 1.21 minutes. In order to account for variations in the length of interactions, we calculated the rate of number of different words by adding all different words produced and dividing by the total time of the observation. Reliability (described below) was based on the number of different words recorded by each transcriber, and was not computed on a word by word basis.

**Control variables—**We used scores from the Leiter-R and the CARS as control variables in the analyses, because both cognitive development and autism symptoms have been linked to differences in vocabulary development (Facon, Facon-Bollengier, & Grubar, 2002; Smith, Mirenda, & Zaidman-Zait, 2007). The Leiter-R is designed for use in populations where traditional cognitive measures may be inadequate. It is normed for participants between the ages of 2 and 20. The Leiter-R is made up of 20 subtests in four domains and yields standardized scores with a mean of 100 and a standard deviation of 15. The present study utilized four subtests, Figure Ground (FG), Form Completion (FC), Sequential Order (SO), and Repeated Patterns (RP), from the Visualization and Reasoning Battery to yield a Brief IQ score. FG requires the individual to find a target image or part of an image in a larger picture. In FC, examinees must "put together" a scrambled image and match it to a target. SO requires examinees to determine what picture comes next in a series. RP has the examinee complete a pattern. We used standard scores from the Leiter-R in our analyses in order to control for both age and developmental differences. Mean scores are presented in Table 1. The lowest standard score of 36 is based on passing at least one item in each of the 4 domains. However, five of our participants passed fewer items (1, 2, or 3). In order to distinguish their scores from those of participants who did pass at least one item in each domain, we used a score of 34 for these 5 individuals. Similar decisions were made in previous research studies (Portoghese et al., 2010; Skinner et al., 2005).

The CARS was used to assess degree of autism symptoms in each child. This 15-item measure yields a general rating of autistic behavior (Mayes et al., 2009). Examiners rate each item on a scale from 1 (within normal limits for age or developmental level) to 4 (severely abnormal for age or developmental level) for different behaviors associated with autism (listening, visual behavior, communication, etc.). The total score can be used to determine where the individual falls on a continuum of autistic behavior. Ratings range from 15 to 60 and can be interpreted as non-autistic (total score of 15 to 29.5), mild or moderately autistic (30–36.5), and severely autistic (37 or higher). Two examiners with extensive

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experience with children with developmental disabilities who had been trained to a criterion of 80% agreement by an experienced researcher obtained CARS scores at each observation. Both research assistants independently completed the CARS. They then compared their scores, discussed any discrepancies and arrived at a consensus about the final score for each item. We obtained some modest variability in scores across the five different observation points. Therefore, for our analyses we used the average CARS scores over the available observations. These average CARS scores and standard deviations are presented in Table 1.

In addition, we considered early maternal responsivity (defined below) as a control variable because we were specifically interested in how sustained responsivity related to vocabulary growth. An earlier study demonstrated that maternal responsivity at our first age period significantly related to language outcomes (Warren et al., 2010). Therefore, we entered this early responsivity into each analysis prior to sustained responsivity in order to determine the added significance of sustained responsivity.

**Maternal responsivity measures—**Maternal responsivity was measured during the mother-child interactions described above. We coded mothers' communication toward their child on a behavior-by-behavior basis using Noldus Observer XT software (Noldus Information Technology, 2008). The coding system was adapted from Landry et al. (1998, 2001) and was described in Warren et al. (2010). This coding system is summarized in the following paragraph.

All maternal 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. Using a multi-tiered coding system, maternal communication acts were coded according to both their level of attention (maintain, introduce, or redirect) and the function of the communication act (request for verbal comply, request for behavioral comply, or comment). Within each tier, these codes were mutually exclusive, and in combination across tiers they were exhaustive. For example, if a mother said, "come over here" while the child was trying to leave the room, it would be coded as a *redirect-request for behavioral comply*. Or, if mother said "that's a big bear" while looking at a book with the child, it would be coded as *maintain-comment*. In addition, the following codes were supplemental in that they could be added to any maternal behavior if observed: gesture, recode (e.g., expansion of a child communication act), communication breakdown (e.g., requesting clarification), or zap (admonishment).

**Coding reliability—**Research assistants completed the primary and reliability coding. Each coder was initially trained to be at least 80% reliable with the first author on a set of videos not part of the current analysis. Following this training, two coders independently coded child and maternal behaviors for each taped observation. After completing independent coding, the two coders compared transcripts, and if agreement was below 80% the coders resolved differences by consensus. This process was implemented to ensure consistency across coders and over time. Approximately 68% files were consensus coded. To determine the inter-judge reliability for the variables *child number of different words* and each behavior that made up the construct of maternal responsivity, we calculated intra-class 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, as well as between primary scores and those arrived at by consensus codes. For child number of different words, the ICC between primary and reliability scores was .97 and between primary and consensus scores was .99. ICCs for maternal behaviors were also high, ranging from .73 and .96 between primary and reliability coders. The ICC of .73 was for maternal recodes, which did not occur as often as other behaviors. ICCs between primary and consensus coders was .99 for all maternal behaviors.

**Measurement schedule—**Five assessment and data collection home visits were completed for each mother-child dyad. Time between the first three assessments was typically 16–18 months. The fourth assessment occurred after an additional round of funding for the project was secured and so the elapsed time between the third and fourth assessment averaged between 30 and 31 months. Time between the fourth and fifth assessment was approximately 18 months. One dyad missed a time 2 assessment; 2 dyads missed a time 4 assessment and 3 dyads missed a time 5 assessment. All of the available data points between child ages 2 and 10 years were used in the growth curve analyses described below.

## Results

The results of this study are organized as follows: first, we describe how the construct of maternal responsivity was derived from our data across multiple data points. Second, we present results of our analyses of the predictive relationships between sustained responsivity and vocabulary outcomes measured with standardized tests. Finally, we present results pertaining to sustained responsivity over time as a predictor of the number of different words produced in mother child interactions across observations using a growth curve analysis.

### Identifying the Maternal Responsivity Component

Maternal responsivity is a complex construct that is indicated by multiple behaviors (Warren & Brady, 2007). Our approach has been to measure behaviors that we hypothesized to be related to the construct of responsivity and then determine mathematically which of the behaviors in combination best represent responsivity. The first step in our analysis, then, was to determine if we could identify a maternal responsivity composite that was comprised of a subset or cluster of the maternal behaviors listed in Table 2. Each of these behaviors are potential indicators of responsivity so we analyzed the data to look for subsets of behaviors that reflect the one underlying construct of interest--responsivity. Preliminary analyses indicated a substantial degree of correlation among the maternal behavior codes. Therefore, we conducted a principal components analysis (PCA;Gorsuch, 2003), which reduced the maternal behaviors to a principal component we labeled maternal responsivity. The PCA uses the correlations among the variables to develop a single component or small set of components that empirically summarizes the correlations among the variables. The component used in this paper was comprised of behaviors coded as *maintain-requests for* *verbal complies*, *maintain-comments*, and *recodes*. We computed z scores for each indicator and then averaged the z scores for each indicator to quantify the amount of responsivity.

For each mother-child dyad, the *early* maternal responsivity score was derived from the first time point after the child was 2 years of age. We then used the average rate of maternal responsivity across observations 2–5 to reflect *sustained* responsivity, because of the substantial amount of variability across mothers over time. Note that the measurement of sustained responsivity was independent of the measurement of early responsivity. Figure 1 shows each mother's maternal responsivity over time. As is shown in this figure, a subset of mothers had relatively low responsivity over all of the observations (*n*=15). Another set of mothers had high responsivity over all observations (*n*=9). A large number of mothers generally increased their rate of responsivity over time (*n*=22), and a final set of mothers were variable—fluctuating between high and low responsivity (*n*=7). Although these patterns of responsivity over time were identifiable, there were insufficient numbers of participants to conduct an analysis of the effects of these different patterns (as was completed in the Landry et al., 2001 study, for example).

Of the three full mutation mothers, two were consistently low in responsivity. One of these actually had the lowest responsivity over time in the whole sample. The third full mutation mother's responsivity was variable with higher-than-average responsivity scores that increased over the first three responsivity observations before decreasing at the fourth observation.

## Responsivity as a Predictor of Vocabulary Outcomes

Prior to conducting regression analyses using the vocabulary variables as outcomes, we calculated Pearson correlations in order to determine the relationships between child control variables, maternal responsivity, the two standardized child vocabulary outcomes, and the number of different words recorded at the same observation as the standardized measures. The results of these correlations are shown in Table 3. Two participants did not have vocabulary data available at age nine and were not included in the regression analyses so the sample size is 53. We provide separate correlations for boys and girls, although it should be noted that the sample of girls was quite small (*n*=10). As the data indicate, all three vocabulary measures were significantly correlated with nonverbal IQ (positively), autism symptom scores on the CARS (negatively), and both early and sustained responsivity (positively). In addition, nonverbal IQ and autism were significantly and negatively correlated with each other. Table 3 also shows the means and standard deviations for each measure. In terms of the vocabulary outcomes for the children of the three full mutation mothers, their raw scores on the PPVT-4 were 40, 58, and 52 (compared to the overall mean of 69.77), and on the EVT-2 were 34, 51, and 60 (compared to the overall mean of 48.06).

**Regression models—**The relationships between both early and sustained maternal responsivity and vocabulary development were further evaluated through two regression models. One model predicted PPVT-4 scores and the second predicted EVT-2 scores when the children were around 8 years old (mean age was 8 years). Fifty-two children passed training items for the PPVT-4 and 44 did so for the EVT-2. A score of 0 was entered for children who did not pass training items. Therefore, the regression models are based on sample sizes of 53. We included both boys and girls in our models to increase variability and sample size. We also provide regression results for a reduced sample including only those participants who passed training items.

To test the hypotheses that maternal responsivity was a significant predictor of both receptive and expressive vocabulary, separate models for the PPVT-4 and EVT-2 outcome variables were constructed. In each model, sequential multiple regression analyses were performed with Leiter IQ scores, maternal education, and average CARS scores entered first as a block. Mean-centered early maternal responsivity was entered second, and mean-centered sustained maternal responsivity was entered as the last step in the modeling. The predictors were mean centered to facilitate interpretation of the coefficients. These models allowed us to determine the degree to which 1) early responsivity predicted later vocabulary outcomes, over and above the control variables entered in the first block, and 2) the degree to which sustained responsivity contributed over and above early responsivity. Tests for multi-collinearity indicated that a low level of multi-collinearity was present for all predictors (tolerance ranged from .42 to .98). See Table 4 for a summary of the regression analyses with PPVT-4 and EVT-2 scores as dependent variables.

As expected, nonverbal IQ and average CARS scores were significantly related to vocabulary outcomes and accounted for variance in outcomes. As shown in Table 4, maternal education was not a significant correlate or predictor. Nevertheless, we left it in the model to account for slight variance that could be associated with maternal education and because maternal education has been significantly related to child IQ in past studies (Mervis, Kistler, John & Morris, 2012). The effect of early responsivity on receptive vocabulary (PPVT-4) scores at age 8 was significant ($p$=.045) when added as the second block, explaining 2% of the variance in PPVT-4 scores. However, when sustained responsivity was added to the model, the effect of early responsivity no longer explained a significant amount of unique variance. Sustained maternal responsivity increased the $R^2$ by another .03 in the PPVT-4 model ($p$=.01).

For expressive vocabulary, early maternal responsivity increased the $R^2$ by less than 1% when EVT-2 was the dependent variable ($p$=.36). The standardized beta for early responsivity before sustained responsivity was added to the model was .08 and nonsignificant. When sustained responsivity was added to the model, the beta for early responsivity remained nonsignificantly different from zero and its value was now slightly negative: −.02.

Sustained maternal responsivity, however, added significantly to the model, increasing $R^2$ by .05 when predicting EVT-2 scores at age 8 ($p$< .01). The final models that included nonverbal cognition, maternal education, autism symptoms and maternal responsivity accounted for 79% of the variance in PPVT-4 scores and 77% of the variance in EVT-2 scores. (See Table 4.) Examination of the part correlations squared in the full model indicates the unique variance accounted for by each predictor. Leiter-R scores, CARS scores, and sustained responsivity scores explained significant unique variance in both PPVT-4 and EVT-2 outcomes. Leiter-R scores uniquely accounted for 4% of the variance in

PPVT-4 and 6% of the variance in EVT-2. CARS scores uniquely accounted for 8% of the variance in PPVT-4 and 5% of the variance in EVT-2. Sustained responsivity uniquely accounted for 3% of the variance in PPVT-4 and 5% of the variance in EVT-2.

In order to determine the effects that scores of zero had on our analyses, we repeated the regression analyses using only those participants who passed the training items (not including zeros for those who did not pass training items). Table 4 includes these results as well. Fifty-two of 53 participants achieved a score on the PPVT-4 and thus the model for PPVT-4 was essentially the same with no changes in significant terms. However, for the EVT-2, the number of cases included dropped from 53 to 42 when scores of 0 were removed. For the smaller sample, autism symptoms (CARS) and sustained responsivity no longer accounted for a significant amount of unique variance and Leiter IQ was the only significant predictor in the full model. Overall, only 66% of the variance in EVT-2 scores was accounted for with the predictors compared to 77% when the full sample was analyzed. The zero-order correlations between EVT-2 scores and sustained responsivity, Leiter IQ, and CARS scores were quite similar in the reduced sample with EVT-2 and Leiter-R: $r$=.77, EVT-2 and Sustained responsivity: $r$ = .45, and EVT-2 and CARS: $r$=−.66. This would suggest that the change in findings was largely the result of power issues associated with the reduction in sample size rather than something inherently different about the participants who did not complete the training items.

**Growth curve models—**In addition to the standardized test scores, we considered the effects of responsivity on the growth of vocabulary measured during mother-child interactions. Previously we reported that early maternal responsivity was a significant predictor of children's number of different words produced when the children were between 11 months and 5 years of age (Warren et al., 2010). For the current analysis we extended the data range and investigated effects of sustained responsivity on vocabulary produced through age 10. Again, we included boys and girls together in the same analysis.

Figure 2 shows the observed rate of different words over time for participating children. Linear and quadratic growth can be observed in the trajectories over time for most children, with more rapid growth occurring at younger ages and a leveling-off of the growth as children aged. There were 7 participants that remained near the floor and spoke very few words throughout the entire course of the study. The rate of different words for the three children with full mutation mothers are shown by solid lines in Figure 2.

Multi-level models were evaluated for rate of different words using SAS PROC MIXED. The observations for each individual in the current data can be viewed as repeated measurements (Level 1) within individuals (Level 2). Age was centered on 8 years so that all intercepts are interpreted as level of communication at 8 years. This age was chosen because it was the same age that the EVT-2 and PPVT-4 outcome measures were obtained for the regression analyses.

Fixed effects terms for intercept, linear growth, and quadratic growth were included in a model with random effects for intercept and slope (linear growth). That is, the trajectory of growth with age was modeled in the fixed effects while allowing individual intercepts and

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slopes to vary (random effects). Restricted maximum likelihood estimation was used when evaluating random effects. Reported parameter estimates are using maximum likelihood. Model comparisons were made using two types of indices: the deviance statistic, or change in the −2 log likelihood, and the Bayesian information criterion (BIC). Only new variables significantly improving the model were retained at each step of the model building. Parameter estimates and probability values for each of the effects are shown in Table 5.

Model 1 is the model with only growth parameters for linear and quadratic growth in rate of different words in the language sample. The intercept value of 5.95 indicates the average rate of different words for participants at 8 years of age. There is significant linear growth over time in rate of different words with average rate increasing by .05 for each month of age. The significant negative quadratic term indicates a slowing in the increasing rate of different words as children get older.

Model 2 includes child predictors (Leiter-R IQ and CARS scores). Both the Leiter-R IQ and CARS scores were significant predictors of the rate of number of different words (words per minute) produced at 8 years of age. The addition of IQ and autism symptom variables significantly improved the model with those having higher Leiter-R scores having higher rates of different words and those with higher CARS scores having lower rates of different words. The next step was to determine if IQ and autism symptoms also significantly influenced rates of change over time as indicated by the linear and quadratic slopes. CARS scores significantly influenced quadratic change over time, and the effect of CARS scores on linear change was also left in the model. The addition of Leiter-R and CARS scores to the model reduced the variance in the intercepts by 49% and reduced the variance of the slopes by 13%.

Model 3 includes the child predictors as well as early and sustained maternal responsivity. Terms for the influence of maternal responsivity on intercepts, linear slope, and quadratic slope were added to the model and evaluated. The results showed that both early and sustained responsivity influenced growth in the number of different words (the slope). Participants who had mothers with high sustained responsivity increased in their rate of different words more quickly, as indicated by the positive parameter estimate. The parameter estimate for early responsivity was negative despite a positive zero-order correlation of $r$=.31 between different words at age 8 and early responsivity. We hypothesize that participants whose mothers were highly responsive early were those who had higher rates of different words at the earliest ages and had slower rates of growth in different words because of ceiling effects in the rate of different words that can be observed in children of this age. Sustained responsivity also significantly influenced the number of different words at 8 years of age with participants whose mothers were high in sustained responsivity having higher rates of different words. The zero-order correlation between sustained responsivity and rate of different words at 8 years was $r$=.65. The predictors included in the models reduced the variance estimate for different words at 8 years from 10.03 with no predictors in the model to 3.21 in Model 3 with all of the predictors included (a 68% reduction in variance). Similarly, the variance estimate for the linear slope was reduced by 53% with the inclusion of the child and maternal predictors to the model.

A graphic depiction of the effect of sustained responsivity obtained from the final model parameters, presented in Figure 3, shows predicted trajectories for rate of different words over time for children who experience relatively high vs. low sustained maternal responsivity. In this model, we used mean levels of child nonverbal IQ and CARS scores, then determined the predicted trajectories for children experiencing mean, high, (1 SD above the mean), or low (1 SD below the mean) sustained responsivity. As can be seen in Figure 3, children whose mothers engage in consistently high rates of responsivity start to separate from the others at around 70 months of age, and by the time the children are 9 years old there are large differences in predicted rate of different words associated with differences in sustained responsivity.

## Discussion

The purpose of this study was to examine the relationship between maternal responsivity and child vocabulary development through middle childhood. Based on our earlier findings, we hypothesized that early maternal responsivity would continue to account for significant variability in child vocabulary outcomes. In addition, we hypothesized that children who experienced high rates of sustained responsivity over time would demonstrate an even greater benefit in terms of vocabulary development. Our results showed that sustained maternal responsivity, represented as average responsivity over times 2–5, significantly added to the regression model for receptive and expressive vocabulary as measured on the PPVT-4 and EVT-2. However, when the participants who received a score of 0 due to not passing training items on the EVT-2 were removed from the analyses, the effects were no longer significant. This may reflect a loss of power and less variability associated with the smaller sample size as the zero-order correlations between the variables were quite similar in the full and reduced samples as noted in the results section.

The growth curve models indicated that sustained maternal responsivity significantly affected growth in the number of different vocabulary words children used during conversational interactions. These results are notable in light of the fact that we controlled for differences in child development, autism symptoms, and maternal education in all of our models. That is, we found that sustained maternal responsivity was a significant predictor of the number of different words children produced at 8 years. Additionally, sustained maternal responsivity significantly influenced *growth* in the number of different words between the ages of 2.9 and 10 years. Children whose mothers demonstrated more sustained responsivity produced higher numbers of different words and displayed higher rates of growth over time. Our analyses applied a rigorous analytic standard by controlling for early responsivity, autism symptoms, nonverbal IQ and maternal education level to determine the unique contribution of sustained responsivity on child vocabulary development.

The results from this study are extend findings by Siller and Sigman (2008) and Landry et al. (2001) who found that early parental responsivity was a significant factor related to language attainments in children during early and middle childhood. As in these earlier studies, we found that children who had mothers who were more responsive, particularly during the early years of our study, had better vocabulary outcomes measured at the middle childhood period. Our findings are also commensurate with those of Landry and colleagues

(Landry, Smith, Swank, & Guttentag, 2008), who found better intervention outcomes when mothers were coached to provide sustained responsivity over both infancy and toddler years. However, the Landry study only followed children through 4 years of age, and our current findings show that sustained responsivity continues to be important through the middle childhood period.

### Implications for intervention

Several intervention implications stem from the current findings. Numerous studies have demonstrated that parents can learn to be more responsive (Fey et al., 2006; Landry et al., 2006; Venker, McDuffie, Ellis Weismer, & Abbeduto, 2011), and this in turn can lead to at least short term improvement in child language (Brady, Warren, & Sterling, 2010; Girolametto, Weitzman, & Clements-Baartman, 1998; Kaiser, Hancock, & Nietfeld, 2000; Reese, Sparks, & Leyva, 2010). However, the long-lasting effects of these interventions remain to be demonstrated. Our data suggest that interventions that enhance parent responsivity during middle childhood, in addition to early childhood, may be beneficial for children with FXS. For example, a program recently developed to target expressive language for 8–10 year old children with expressive language delays found that parent implemented interventions improved conversational skills (Allen & Marshall, 2011). However, there is much yet to learn about appropriate contexts and expectations for interventions targeting parent-child interactions during the middle childhood years.

### Variability over time

One area that needs further consideration is the conceptualization of optimal parent-child interactions as children age. In the current study, we faced the challenge of measuring responsivity over many years and found that the topography of responsivity changed as children advanced from early to middle childhood. For example, providing many comments and gestures was indicative of responsivity early on (Warren et al., 2010), but less important in the current study when children moved into middle childhood and became more independent communicators. Our principal components analyses did find that *requests for verbal comply* (e.g., “What’s that?”), *comment* (e.g., that’s a big cookie”), and *recode* (“oh you’re telling me you want more”) continued to be consistent indicators of responsivity from early to middle childhood. Further research is needed to determine the extent to which these indicators continue to represent responsivity, or are replaced by other parent behaviors that are more related to communication during later childhood and adolescence.

The longitudinal data collected for this study allowed us to identify different patterns of responsivity, with some mothers showing consistently high responsivity, some varying widely from observation to observation, and a few who were consistently low. Although it was not our purpose to investigate what factors contribute to these different patterns, it may be that changes in family supports and personal life stressors contribute to responsivity measured on a given day or as both a parent and child age. That is, as children move into adolescence, changes in their behaviors may impact how parents respond to or engage them. Likewise, there is some evidence that carrier mothers may become more susceptible to stress and other variables as they age (Abbeduto et al., 2004).

Certain maternal variables, such as low educational attainment (i.e., less than a high school education), mild intellectual disability 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 (with the exception of the three full mutation mothers), 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 include an increasing risk of cognitive deficits in attention, verbal memory, and executive function as the mothers age (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, Rogeness, McClure, Clayton, & Johnson, 1996). These risk factors have been associated with lower maternal responsivity (Goldsmith & Rogoff, 1995; Osofsky & Thompson, 2000).

In addition, changes in maternal responsivity may be due to child changes over time. That is, maternal responsivity not only contributes to child development but also may be impacted by the lack of changes in child development (Sterling, Warren, Brady, & Fleming, in press). Hence, an additional challenge is evaluating the contribution of responsivity to children who are developing at different rates due to variations associated with the fragile X phenotype. Ideally, one would analyze the mutual effects between caregiver and child using a time-varying covariates approach, but such an analysis would require larger numbers of participants than we have available.

## Study limitations

As previously noted, the study has several limitations. These include a relatively small sample size and the use of a sample of convenience over a randomly selected group. In addition, we focused only on vocabulary as opposed to more comprehensive measures of language and cognitive development. Future studies may wish to broaden the scope of the outcome measures. Finally, we did not evaluate any effects that could be associated with a negative as opposed to responsive interaction style. The decision to only focus on positive parenting (responsivity) was based on earlier findings that did not demonstrate any link between more negative interactions and child outcomes in our data. We did observe a few instances of negative behaviors such as admonishments or verbal restrictions during our observations. Increases in these types of maternal responses may be seen as children get older and sometimes demonstrate more undesirable behaviors. Further study at these older ages may in fact show increases in both disruptive child behaviors and parenting styles that attempt to mitigate such behaviors.

## Conclusions

The way in which communicative partners, particularly mothers, interact with their children is a powerful variable in children's language development. Our findings contribute to the growing research base demonstrating that responsivity can have long-lasting impact. Importantly, we showed that children who experience consistent sustained responsivity have better vocabulary outcomes in middle childhood. Further research is needed to determine how best to capitalize on these results in terms of developing treatments that increase responsivity by parents and other partners, such as teachers and peers. Increasing responsivity appears likely to provide added benefit to children with FXS receiving speech and language interventions.

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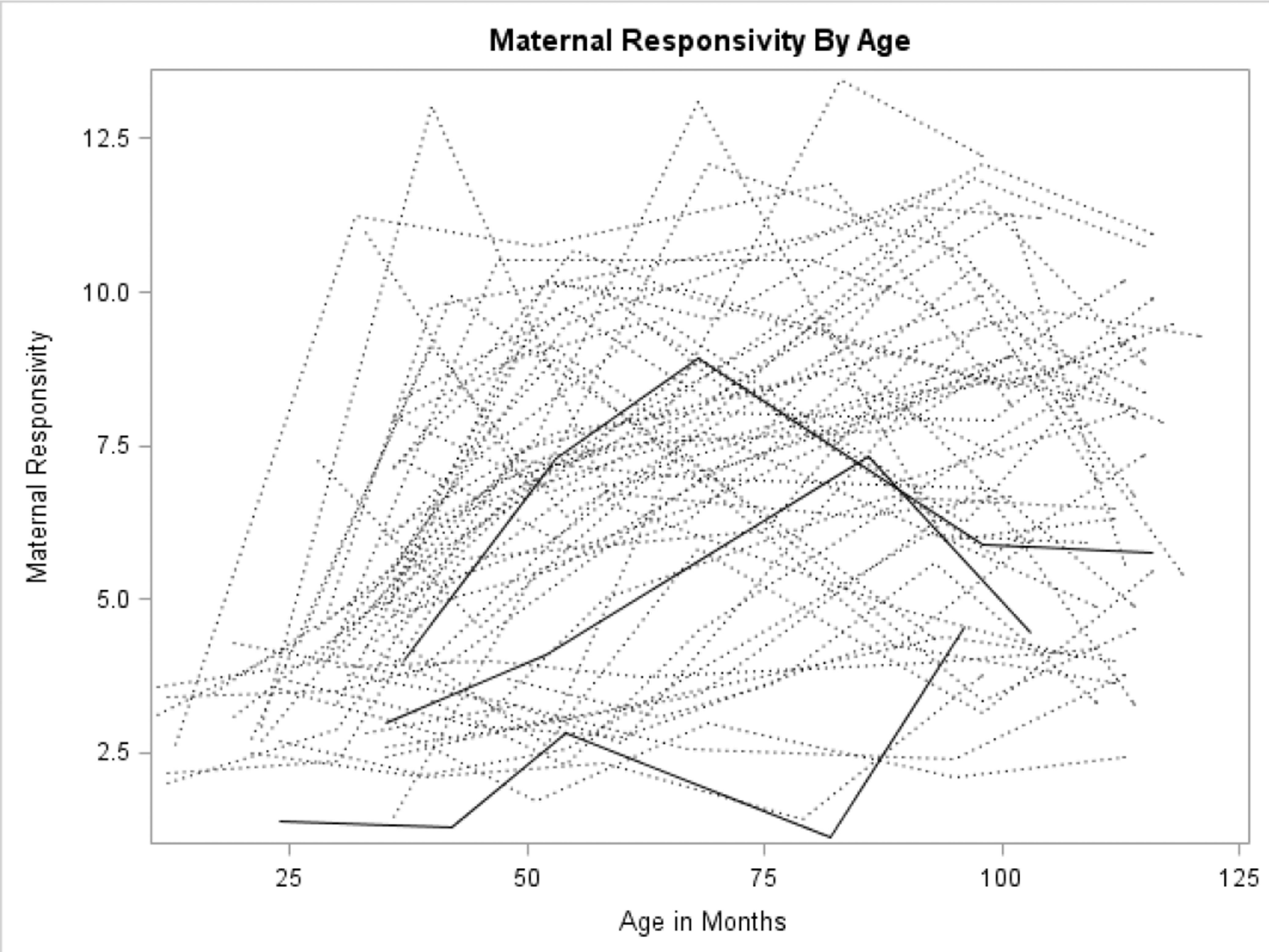


**Figure 1.**
Maternal responsivity over time. Solid lines are Full Mutation mothers.

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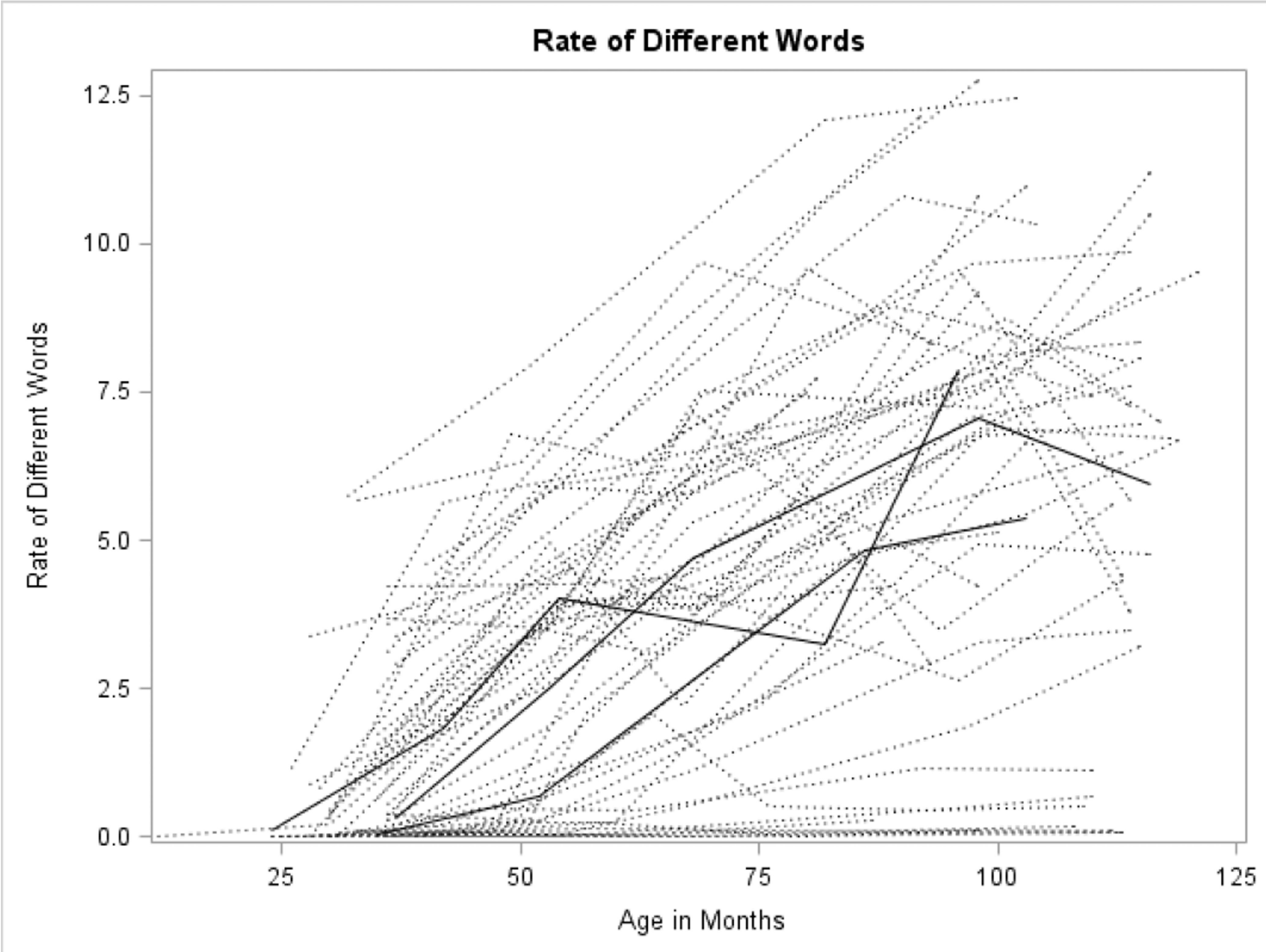


**Figure 2.**
Rate of different words over time. Children whose mothers are Full Mutation are in solid lines.

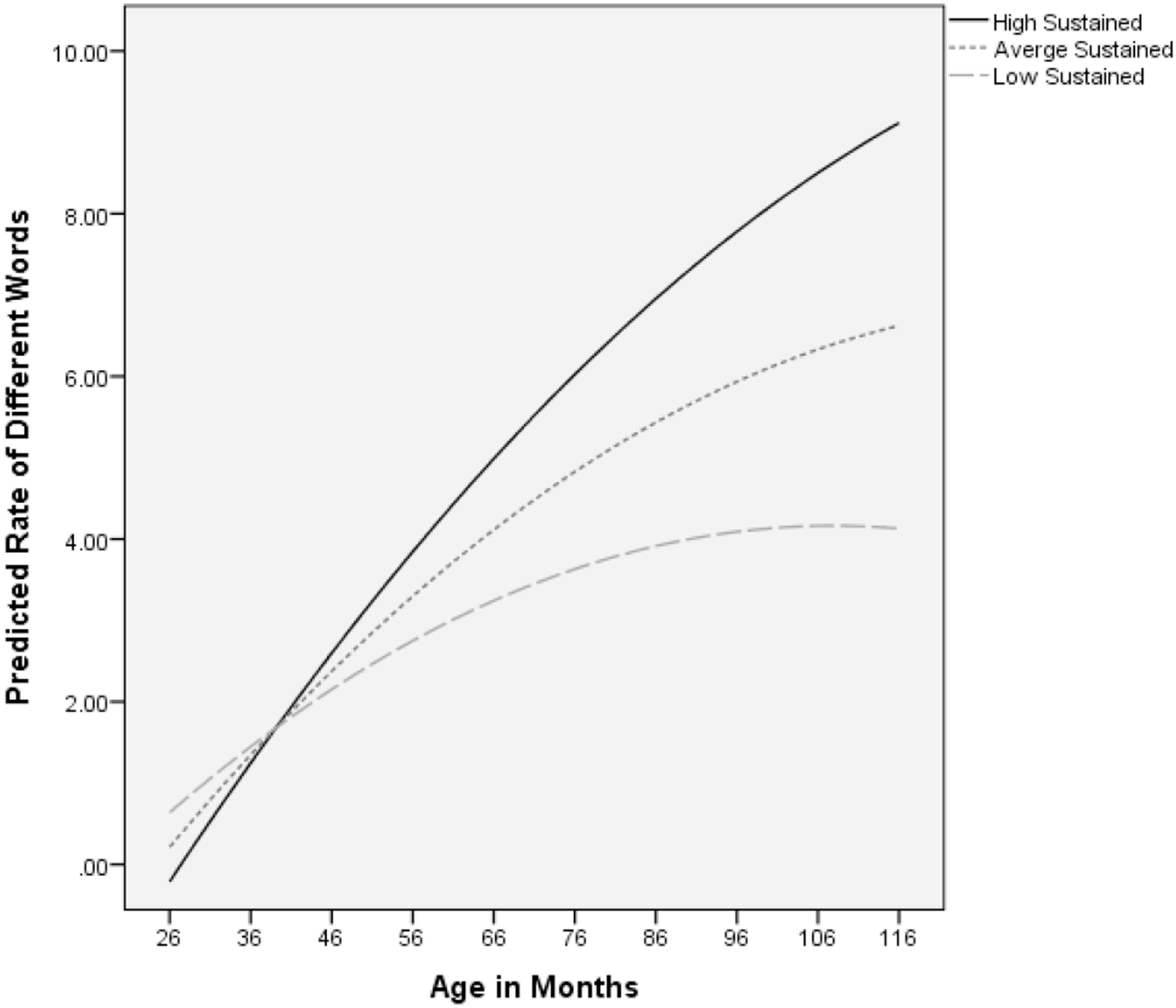


**Figure 3.**

Predicted trajectories of growth in number of different words for children with high, average, or low sustained responsivity*.

*Predicted scores for participants with average Leiter-R, CARS, and Early Responsivity Scores and scores at the mean or one standard deviation above or below the mean on Sustained Responsivity.

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**Table 1**

Child Descriptive Information and Scores from the PPVT-4/EVT-2.

| Variable | *M* | *SD* | Range |
|---|---|---|---|
| Chronological Age [a] | 8.06 | 0.37 | 7;3 – 8;8 |
| Nonverbal IQ[b] | 52.26 | 15.61 | 34–98 |
| CARS score[c] | 26.60 | 5.48 | 16.60–37.5 |
| PPVT-4[d] | 71.12 | 35.22 | 3–143 |
| EVT-2[e] | 48.98 | 31.34 | 0–114 |

[a,b]based on the Leiter-R International Performance Scale-Revised

[c]Average scores from the Childhood Autism Rating Scale

[d]Peabody Picture Vocabulary Test, 4th edition

[e]Expressive Vocabulary Test, 2nd edition

**Table 2**

Definitions of Maternal Behaviors

| **Category & Behavior** | **Definition** | **Example** |
|---|---|---|
| Level of Attention | | |
| Maintain | Mom references toy, behavior, or emotional state of the child | Child is playing with a doll and mom says "You're feeding the baby." |
| Redirect | Child is actively involved physically and/or visually in play with object different from the one mom presents or references | Child is playing with a toy and mom says, "What else do you want to play with?" |
| Introduce | Mom presents a new object or activity at a time child is not actively attending to anything | Child is gazing into space and mom says, "Let's get a snack." |
| Reading | Mom reads a book verbatim without comment, further explanation, or description | Mom reads a book aloud, "Once upon a time…" |
| Function | | |
| Request for verbal comply | Questions or statements intended to elicit a verbal response | Mom says, "say ___" or "ok?" at the end of a comment |
| Request for Behavioral comply | Directives to which the child can comply behaviorally | Mom says, "Push this one," or "I want you to do it." |
| Comment | All comments | Mom says "Good job!" or "That's the blue block" |
| Supplemental | | |
| Gesture | Non-verbal idea transmission through conventional or unconventional gestures and sign language | Mom points to a book and says "Do you want to read this?" |
| Recode | Verbal interpretation of child's communication act | Child says "ba" and mom says, "Do you want your ball?" |
| Communication breakdown | Mom asks for clarification of child's communication or gives no verbal or nonverbal response when one is called for | Child mumbles and mom says, "What did you say?" |
| Zap | Verbal or non-verbal restriction of child's behavior | Mom says "Don't do that" and shakes her finger at the child |

## Table 3

Correlations between Measures and Summary Statistics for Males and Females

| | *1* | *2* | *3* | *4* | *5* | *6* | *7* | *8* | $M_{girls}$ | $SD_{girls}$ |
|---|---|---|---|---|---|---|---|---|---|---|
| 1. PPVT-4 | - | .96** | .93** | −.13 | −.75** | .72** | .61* | .52 | 93.60 | 44.52 |
| 2. EVT-2 | .74** | - | .95** | −.24 | −.75** | .59* | .54 | .62 | 69.20 | 39.50 |
| 3. Leiter-R | .65** | .66** | - | −.23 | −.83** | .58* | .43 | .42 | 68.20 | 21.16 |
| 4. Maternal Ed[a] | .07 | .03 | .03* | - | .51 | .06 | .15 | −.43 | 14.70 | 1.89 |
| 5. CARS | −.83** | −.68** | −.68** | −.11 | - | −.51 | −.12 | −.40 | 23.38 | .10 |
| 6. Early Resp[b] | .35* | .20 | .20* | .04 | −.29* | - | .73** | .63 | −.10 | 2.92 |
| 7. Sustained Resp[b] | .72** | .65** | .65** | .05 | −.70** | .39** | - | .54 | .23 | 2.67 |
| 8. Rate of DW[c] | .77** | .82** | .60** | −.14 | −.72** | .24 | .75** | - | 7.06 | 3.84 |
| $M_{Boys}$ | 64.23 | 44.17 | 48.56 | 15.65 | 27.35 | −.13 | −.05 | 5.93 | | |
| $SD_{Boys}$ | 32.14 | 27.49 | 11.46 | 2.41 | 5.04 | 2.24 | 2.31 | 3.58 | | |

*Note*. Intercorrelations for males (N=43) are presented below the diagonal, and intercorrelations for females (N=10) are presented above the diagonal. Means and standard deviations for males are presented in the horizontal rows, and means and standard deviations for females are presented in the vertical columns.

** Correlation is significant at the 0.01 level (2-tailed)

* Correlation is significant at the 0.05 level (2-tailed)

[a] Education

[b] Responsivity

[c] Different Words

## Table 4

Sequential Multiple Regression Analyses Predicting PPVT-4 and EVT-2 Scores

| **Predictor** | **PPVT-4 Full** | | **EVT-2 Full** | |
|---|---|---|---|---|
| | **Δ $R^2$** | **β** | **Δ $R^2$** | **β** |
| Step 1 | .74*** | | .71*** | |
| Maternal Education | | .01 | | −.03 |
| Leiter Nonverbal IQ | | .29** | | .38** |
| Average CARS | | −.46** | | −.34** |
| Step 2 | .02* | | .01 | |
| Early Responsivity | | .09 | | −.02 |
| Step 3 | .03* | | .05** | |
| Sustained Responsivity | | .23* | | .30** |
| Total $R^2$ | .79** | | .77** | |
| *N* | 53 | | 53 | |

| Predictor | PPVT-4 no 0's | | EVT-2 no 0's | |
|---|---|---|---|---|
| | Δ $R^2$ | β | Δ $R^2$ | β |
| Step 1 | .73*** | | .62*** | |
| Maternal Education | | .05 | | .04 |
| Leiter Nonverbal IQ | | .37** | | .60** |
| Average CARS | | −.55** | | −.23 |
| Step 2 | .02 | | .03 | |
| Early Responsivity | | .14 | | .19 |
| Step 3 | .03* | | .01 | |
| Sustained Responsivity | | .22* | | .14 |
| Total $R^2$ | .77** | | .66** | |
| *n* | 52 | | 44 | |

* $p < .05$.

** $p < .01$. Betas are from full model with all predictors.

## Table 5

Fixed Effect Estimates (Top) and Variance-Covariance Estimates (Bottom) for Rate of Number of Different Words Models with Child and Maternal Predictors

| Parameter | Model 1 | Model 2 | Model 3 |
|---|---|---|---|
| | | Fixed Effects | |
| Intercept | 5.95 (.44)*** | 12.52 (2.30)*** | 8.88 (2.02)*** |
| Level 1 | | | |
| Linear | .05 (.01)*** | .06 (.04) | .01 (.04) |
| Quadratic | −.001 (.0001)*** | −.002 (.0006)** | −.002 (.001)*** |
| Level 2 | | | |
| Leiter-R IQ on Intercept | | .04 (.02)* | .04 (.01)** |
| CARS on Intercept | | −.33 (.07)*** | −.18 (.06)** |
| CARS on Linear | | −.0005 (.001) | .001 (.001) |
| CARS on Quadratic | | .00005 (.00002)* | .0001 (.00002)** |
| Predictor Variables | | | |
| Early Maternal Responsivity on Intercept | | | −.07 (.14) |
| Early Maternal Responsivity on Linear | | | −.007 (.002)** |
| Sustained Maternal Responsivity on Intercept | | | .83 (.13)*** |
| Sustained Maternal Responsivity on Linear | | | .01 (.002)*** |
| | | Random Parameters | |
| Intercept | 10.03 (2.03)*** | 5.08 (1.10)*** | 2.55 (.61)*** |
| Slope | .002 (.0004)*** | .001 (.0003)*** | .001 (.0002)*** |
| Covariance | .11 (.03)*** | .07 (.02)*** | .05 (.01)*** |
| Residual | 1.32 (.16)*** | 1.29 (.16)*** | 1.31 (.16) *** |
| -2 log likelihood | 1007.6 | 938.7 | 893.8 |
| BIC | 1035.6 | 982.6 | 953.6 |