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Maternal Well-Being and Child Behavior in Families with Fragile X Syndrome

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Abstract

The purpose of this study was to examine the bidirectional relationshipsnetres behavioral functioning of children with fragile X syndrome (FXS), the leading cause of inherited intellectual disability. Children with FXS commonly demonstrate challenging behavior related to anxiety, attention, and aggression, whereas mothers of children with FXS have been identified as susceptible to mental health disorders due to their status as genetic carriers of the FXS premutation, as well as the environmental stressors of raising children with special needs. The longitudinal design of this study builds upon prior work that established a concurrent relationship among these factors in families of children with other intellectual disorders. Findings indicated that maternal mental health status was not significantly related to changes in levels of child challenging behavior, child challenging behavior was related to changes in maternal depression over time, and heightened levels of child challenging behavior was related to increased feelings of maternal closeness toward the child over time. The unexpected nature of the result regarding maternal closeness provides new and more complex hypotheses about how mothers of special needs children demonstrate adaptation and resilience. The findings have implications for maternal and familial mental health treatment as well as future research.

Keywords

fragile X syndrome; mental health; maternal outcomes; child behavior

1.1 Introduction

Fragile X syndrome (FXS) is the leading cause of inherited intellectual disability (Hagerman 2008). The syndrome results from an expansion of a trinucleotide (CGG) sequence in the *FMR1* gene on the X chromosome, which leads to a deficit of FMRP, a protein that is essential for normal neural functioning (Bassell & Warren, 2008). In the full mutation case, which produces FXS, the CGG sequence is expanded to more than 200 repetitions compared

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to the healthy allele range of 15 to 54 repetitions (Brown, 2002). In the premutation case, the expansion is between 55 and 200 repetitions (Brown, 2002). The premutation can result in both reduced FMRP levels and elevated levels of *FMR1* messenger RNA and possible RNA toxicity (Tassone et al., 2000). Although the premutation does not produce FXS, the premutation is associated with adverse phenotypic consequences, including comorbid conditions, such as the neurodegenerative disorder FXTAS (Cornish, Turk, & Hagerman, 2008). In virtually all cases, FXS is inherited from the mother, who will be a carrier of either the *FMR1* premutation or full mutation (Nolin et al., 1996). Thus, FXS is a multigenerational disorder and the functioning of each family member is likely to be affected by, and affect, the functioning of other family members (Seltzer, Abbeduto, Greenberg, Almeida, Hong, and Witt, 2009). The aim of the present study was to characterize the dynamic bidirectional relationships that exist among child, mother, and family context over time in families affected by FXS.

1.1.1 Phenotype of Children and Youth with FXS

Most males with FXS have a moderate intellectual disability, and although females are less impaired on average, up to half also meet criteria for an intellectual disability (Hagerman & Hagerman, 2002). In addition to intellectual impairments, FXS is associated with an elevated rate of challenging behaviors relative to conditions such as Down syndrome, although there is considerable within-syndrome variability in this regard (Kau et al., 2004). Behavioral problems associated with FXS include social anxiety, hyperactivity, hypersensitivity to sensory stimuli, increased aggression, self-injurious behaviors, and attention problems (Kau et al., 2004; Kaufmann et al., 2004). In addition, individuals with FXS commonly have impaired social and communicative skills (Abbeduto et al., 2007; Lewis et al., 2006) and other behaviors characteristic of autism (Bailey, Sideris, McDuffie et al., 2012; Roberts, & Hatton 2008). In fact, 25% to 33% of people with FXS meet criteria for a co-morbid diagnosis of autistic disorder, with the remainder displaying at least some autistic-like behaviors (Bailey et al., 2004; Brown et al., 1982; Demark et al., 2003; Lewis et al., 2006; Rogers et al., 2001). For males with FXS, a co-morbid autism diagnosis or more severe autism symptoms are associated with more problem and aberrant behavior (especially social avoidance and repetitive behavior), lower levels of adaptive behavior, more severe language impairments, and lower IQ scores relative to boys with FXS alone (Kau et al., 2004; McDuffie et al., 2010; McDuffie et al., 2012).

There is considerable evidence from studies of children with intellectual disabilities of various origins that child challenging behavior is a powerful predictor of maternal stress and poor mental health (Abbeduto et al., 2004; Roberts et al., 2009; Wheeler et al., 2010). At the same time, there is evidence that the child's challenging behaviors can be exacerbated by increased maternal stress and mental health problems, such as depression (Jouriles et al., 1989; NICHD 1999; Orsmond, Seltzer, Krauss & Hong 2003; Osofsky & Thompson 2000). In the present study, we were interested in the ways in which the challenging behavior of the son or daughter with FXS affects, and is affected by, the mental health of the mother as well as the family climate.

1.1.2 Maternal Mental Health

Biological mothers of individuals with FXS are at elevated risk for mental health concerns. In a sample of these mothers who had clinically significant levels of stress according to self-report, 63% also exceeded the clinical threshold on at least one other measure of maladaptive mental health (Bailey et al., 2008). Relative to women in the general population, biological mothers of children with FXS also display higher rates of social phobia, personality disorders (especially schizotypal personality disorder), major depressive disorder, panic disorder, and agoraphobia (Franke et al., 1998; Roberts et al., 2009). Depression and anxiety, however, are the most frequently diagnosed psychiatric disorders for women with the *FMR1* premutation (Franke et al., 1996). Rates of depression for women with the premutation have been identified as ranging from 16% to 40% (Bailey et al., 2008; Franke et al., 1996). Lifetime rates of depression have been cited at 56%, which is far higher than the 10 to 12% of women who experience depression in the general population (Wheeler et al., 2007). The rate of current diagnoses of anxiety disorders in female carriers of the *FMR1* premutation has been found to be 17% (Bailey et al., 2008), and mothers of children with FXS have a frequency of anxiety disorders that is three times higher than that of mothers of children with autism and children who are typically developing (Franke et al., 1996).

It has been suggested that biological mothers of individuals with FXS are more susceptible to mental health problems in part because of their own genetic status as carriers of either the *FMR1* full mutation or premutation (Roberts et al., 2009). The evidence on this point, however, is equivocal. Thompson et al., (1996) found that mothers of individuals with FXS had a higher rate of depression (78%) than mothers of children with Down syndrome or spina bifida (37%) who, they argued, cope with similar environmental stressors. In fact, the rates of most types of challenging behaviors, including those reflective of externalizing problems, are higher in FXS than in Down syndrome, spina bifida, and many other syndromes (Dykens et al., 2000); thus, the Thompson et al. data are ambiguous as to the cause of maternal differences in mental health. In addition, Roberts et al. (2009) found that the age-of-onset for psychiatric diagnoses in mothers who were *FMR1* expansion carriers occurred much earlier than did their child's diagnosis, which led these investigators to suggest that the high prevalence of affective disorders among the mothers could not be attributed solely to the stress of raising a child with a developmental disability. However, Bailey et al. (2009) found that the developmental problems of children with FXS are manifested and recognized by parents years in advance of the FXS diagnosis, which raises the possibility that child behavior and delays prior to diagnosis might still be contributing to maternal mental health problems. Nevertheless, relatives of mothers of children with FXS also have a higher frequency of affective disorders (20%) compared to the relatives of mothers of children with autism (11.7%) and relatives of mothers from the general population (3.3%; Franke et al., 1996), supporting the claim that being a premutation carrier confers risk for mental health problems in and of itself. It is likely, however, that it is the dual action of a genetic predisposition and the experiences associated with parenting a son or daughter with severe behavioral challenges that leads to less positive maternal mental health outcomes.

Indeed, there have been suggestions that the stress arising from the parenting role contributes to reduced well-being among mothers of individuals with FXS. Roberts et al. (2009) found that the presence of anxiety disorders in mothers with the *FMR1* premutation was not predicted by genetic variables, but was instead strongly related to child variables. In particular, mothers of multiple children with FXS who had elevated scores on the Child Behavior Checklist also displayed higher levels of anxiety (Roberts et al., 2009). Similarly, Abbeduto et al. (2004) found that the strongest predictor of maternal depressive symptoms was the extent and severity of the affected child's behavioral symptoms, a finding replicated by Bailey, Sideris, Roberts, and Hatton (2008). In another study of families of children with FXS by Wheeler et al. (2010), child autistic behaviors, challenging behavior, and lack of adaptability were found to be highly correlated with parenting stress, which was itself correlated with maternal depression. Wheeler et al. (2007) found that 55% of the mothers of children with FXS in their sample who reported experiencing significant levels of stress attributed this emotional strain to difficult child behavior. In all of these studies, however, only concurrent relations between maternal psychological status and child behavior problems were examined, which makes it impossible to examine an ongoing relationship between the variables. An exception is Hartley et al. (2012), who found that child challenging behavior could affect subsequent maternal physiological stress, at least for a subset of women carrying the *FMR1* premutation; however, these investigators examined these relationships only over a 24-hour period, leaving longer-term relationship unexplored. Moreover, mothers have frequently been the reporters on both their own mental health and the challenging behaviors of their children, raising the possibility that the relationships observed reflect wholly or in part a reporter bias. There is, then, a need to clarify the relationship between maternal mental health and child problem behavior over the course of development in families affected by FXS. In the present study, we used a short-term longitudinal design to examine the extent to which child challenging behavior leads to change over time in maternal mental health. Additionally, fathers rather than mothers were asked to report on child challenging behavior so as to minimize the potential effects of a reporter bias.

Maternal mental health can influence, and be influenced by, the behaviors and characteristics of the child. In fact, there is abundant evidence that maternal mental health problems can have detrimental effects on child social, linguistic, behavioral, and scholastic outcomes (NICHD, 1999). Children of chronically depressed mothers have poorer functioning in the domains of school readiness, verbal comprehension, expressive language, cooperation, and behavior (NICHD 1999). Mothers who are depressed and therefore demonstrate little sensitivity in their parent-child interactions are more likely to have children who exhibit problematic behavior (NICHD, 1999; Wheeler et al., 2010). Poor maternal mental health status lowers the level of maternal engagement (Wheeler et al., 2007), responsivity to the child (Osofsky & Thompson 2000), sensitivity within the parent-child relationship (NICHD, 1999), and the occurrence of positive parent-child interactions (Jouriles et al., 1989). Such negative effects appear to be mediated by the nature of the maternal behaviors directed toward the child.

In contrast, developmental outcomes for children with disabilities are improved when parents establish a caring, supportive, and positive family environment (Mink & Nihira, 1986). Children with disabilities who experience cohesion, harmony, and an expressive and child-oriented family environment have been found to have higher levels of adaptive behavior, fewer behavior problems, and experience less peer isolation (Mink et al., 1983). Highly responsive parenting also is related to more positive language outcomes for children with developmental disabilities, including those with FXS (Warren et al., 2010). Conversely, there is evidence from studies of families of adults with intellectual disabilities of varying etiology that declines in maternal psychological well-being can exacerbate child challenging behavior over time (Orsmond et al., 2003). In the present study, we evaluated the extent to which this relationship between changes in maternal mental health and child behavior is also characteristic of families affected by FXS.

1.1.3 Pessimism, Closeness, and the Family Environment

Maternal perceptions of the family and relationships within the family may also be impacted by FXS. For example, compared to mothers of adolescents with other developmental disabilities (e.g. Down syndrome), mothers of adolescents and young adults with FXS are more pessimistic about their child's future (Abbeduto et al., 2004; Lewis et al., 2006). High levels of parental pessimism can be problematic because they can contribute to parental stress and serve as a barrier to positive child outcomes. Also, parents who are pessimistic about their child's future level of functioning may be less likely to utilize available services, possibly because they are not hopeful that such services will benefit their child (Floyd & Gallagher, 1997).

Closeness within the mother-child relationship is another domain within the family environment that can be impacted in families of individuals with FXS. Mothers of adolescents and young adults with FXS perceive less reciprocated closeness with their children than do mothers of adolescents with Down syndrome (Abbeduto et al., 2004). Some of the social and emotional characteristics (e.g. eye gaze aversion) of children with FXS may contribute to the difference between FXS and other developmental disabilities in terms of maternal perceptions of closeness. In fact, mothers of sons with co-morbid FXS and autism report perceiving that their children feel less close toward them than do mothers of sons who have only FXS (Lewis et al., 2006).

Conflict within the family environment also can be impacted by having a child with FXS. Mothers of sons with FXS (with and without co-morbid autism) have reported higher levels of family conflict than mothers of children with Down syndrome (Lewis et al., 2006). Family conflict may interfere with the warm and positive parental engagement that characterizes cohesiveness within the family, which has been identified as positively influencing children's emotional and behavioral outcomes (Eisenberg et al., 2005). Indeed, higher levels of family cohesion combined with high levels of parent involvement have been found to result in children with various disabilities demonstrating better independent functioning and social awareness (Mink & Nihira 1986).

The negative impact of the characteristics associated with FXS on the family environment may be due to poor maternal mental health, child challenging behavior, or both. Abbeduto et

al. (2004) found that maladaptive child behavior predicted higher levels of maternal pessimism and less closeness in the mother-child relationship. Children's autistic behaviors also have been found to be significantly correlated with negative control behaviors by mothers (Wheeler et al., 2010). In contrast, children with FXS display less task-related frustration if their mothers are more responsive (Wheeler et al., 2010). Although suggestive, the relationships observed in these studies have emerged from concurrently measured variables, leaving the direction of this connection unclear.

In summary, it is clear that levels of conflict, cohesion, and pessimism have important potential implications for the entire family system. Studies have established that concurrent relationships exist between these variables and aspects of child challenging behavior and maternal mental health; however, longitudinal studies are needed to disentangle the relationships among these variables. The present study was focused on the short-term longitudinal relationships among the behavioral characteristics of children with FXS, maternal mental health problems, and the family environment.

1.1.4 The Family as a System

Family systems theory suggests that the trends identified regarding maternal mental health, child behavior, and family environment for families with FXS are significant because all three aspects of family functioning have the potential to influence one another (von Bertalanffy, 1968). Because familial relationships exist as a system, the experience of one family member affects all members of the family in some way (Minuchin, 1974). This framework provides a rationale for examining the bidirectional relationship between maternal mental health status and child behavioral outcomes, as well as for examining the effects of both of these factors on the overall family environment. Such data are critical for identifying the factors that affect the family system most powerfully and that should be targeted by clinical intervention.

1.1.5 The Present Study

It is clear that the elevated rate of mental health problems for mothers of children with FXS can create barriers to well-being for mothers, children, and families. What is still unclear is how factors within the family system, specifically maternal mental health status, challenging child behavior, and the family environment, influence changes in one another over time. The present study was designed to establish directionality among these constructs. Knowledge of how these factors influence one another can aid in identifying families that are at risk and can help direct clinical interventions with mothers, affected children, and the entire family system. Thus, we addressed the following research questions:

1. Does maternal mental health status predict change in child challenging behavior?
2. Does child challenging behavior predict change in maternal mental health status?
3. Do maternal mental health status and child challenging behavior predict change in the family environment, particularly in levels of family cohesion, family conflict, and maternal pessimism regarding the future of the child with FXS?

1.2 Material and Methods

The present sample was comprised of 18 biological mothers of youth who had a genetic diagnosis of FXS. If a family had two children with qualifying characteristics, only one sibling (randomly determined) was included in the present analyses.

1.2.1 Participants

Recruitment—Families participating in a larger ongoing study of language learning in children and adolescents with FXS were invited to participate. Additional participants were recruited through newsletters, websites of national developmental disabilities organizations, and the national and regional chapters of disability advocacy organizations. Participants were also identified through the use of a university research registry. The participating families lived in 11 different states in the U.S.

Confirmation of fragile X syndrome—Genetic reports were requested from mothers to confirm FXS in their biological children. Of the 18 mothers in our sample, 15 carried the *FMR1* premutation, two had mosaicism (i.e., full and premutation), and one could not provide genetic test results. Within our sample of children, 12 had the FXS full mutation, three had mosaicism, and genetic test results were unavailable for three children, although other reports for these children corroborated maternal report of the diagnosis.

Inclusionary criteria for the current analysis—This study was designed to examine changes in child behavior, family environment, and maternal mental health across two time points. The target length of time between time points one (T1) and two (T2) was one year, but flexibility in this year-long window was necessary to allow for scheduling conflicts, family emergencies, illnesses, etc. The length of time allowed for inclusion between T1 and T2 ranged from 294 to 436 days. The mean number of days between T1 and T2 was 387.9, with a standard deviation of 31.6.

Inclusion in the current analyses also required that participants had completed relevant maternal mental health measures (SCID & SCL-90-R, described below) and paternal reports of child behavior (CBCL, ABC, and PBS, described below). Due to the importance of paternal reports in our analyses, being a member of a two-parent household was a requirement for inclusion in the present study.

1.2.2 Measures

Maternal mental health status was evaluated using three instruments. First, the Structured Clinical Interview for DSM-IV-Axis I Disorders/Non-patient research version (SCID; First et al., 2002) was used to identify symptoms of Axis I disorders as defined in the DSM-IV. This assessment was administered to each mother at T1 only, by a graduate student clinician who had completed training coordinated by Robert First, one of the developers of the SCID. Inter-rater agreement was determined by having a licensed clinical psychologist evaluate 25% of the audiorecorded SCID samples administered, which included more than just the mothers in the present analysis. Agreement regarding administration fidelity was 88%. There was an 86% consensus for determining if a psychiatric diagnosis was warranted. Of

the cases in which a psychiatric diagnosis was assigned, agreement was 100% regarding the specific diagnosis (or diagnoses) thought appropriate. For analysis, we used a binary classification in which each mother received a score of 1 if she met criteria for one or more lifetime psychiatric diagnoses and a 0 otherwise. Eleven mothers met criteria for at least one lifetime diagnosis.

The Symptom Checklist-90-R (SCL-90-R; Derogatis 1993) was completed by each mother to assess the presence of 90 symptoms of psychopathology. Domains of psychopathology measured in this assessment are Somatization, Obsessive-Compulsive, Interpersonal Sensitivity, Depression, Anxiety, Hostility, Phobic Anxiety, Paranoid Ideation, Psychoticism, and additional miscellaneous items. The SCL-90-R is a self-report measure. We used the Global Severity Index (GSI) score from this measure, which provides a summary of symptoms across all domains, with higher scores reflecting more mental health concerns. Cronbach's alpha coefficients for internal consistency for the individual subscales of this measure ranged from .71 (Obsessive-Compulsive Subscale) to .93 (Depression subscale) at T1 and .79 (Hostility subscale) to .95 (Depression subscale) at T2. (Note that a coefficient could not be computed for Phobic Anxiety at T1 because virtually no one endorsed any of its items; therefore, most responses were zeros.) The mean GSI scores for the sample were 54.4 and 53.0 for T1 and T2, respectively.

Symptoms of maternal depression were also measured via self-report using the Center for Epidemiologic Studies Depression Scale (CES-D; Radloff 1977). This scale assesses the frequency of 20 symptoms of depressed mood or affect on a 4-point Likert scale with reference to the past week. Higher scores reflect more symptoms or more severe symptoms. Cronbach's alpha reliability was 0.72 at T1 and 0.75 at T2 for this measure. The means for the sample were 9.6 and 11.2 at T1 and T2, respectively.

Closeness in the mother-child relationship was measured using the Positive Affect Index (PAI; Bengtson & Black 1973), which was completed by mothers. This 10-item self-report scale assesses the degree of understanding, trust, fairness, respect, and affection that the mother feels toward her child and the mother's perception of the child's reciprocated closeness. Items are rated on a six-point scale, with higher ratings indicating better quality of relationship. For this sample, Cronbach's alpha reliability for maternal feelings toward the child items was 0.71 at T1 and 0.70 at T2. Cronbach's alpha reliability for perceived reciprocated closeness from the child was 0.68 at T1 and 0.83 at T2. The means for maternal feelings of closeness toward the child were 26.4, and the means for maternal perception of child-reciprocated closeness were 25.5, at both T1 and T2.

The pessimism subscale from the Questionnaire on Resources and Stress (QRS-F; Friedrich, Greenberg & Crnic 1983) was used to assess maternal pessimism about her child's prospects of achieving future self-sufficiency. This 11-item, self-report measure requires mothers to rate each item as true or false, with higher scores indicating more pessimism. Cronbach's alpha reliability for our sample was 0.64 at T1 and 0.73 at T2. The means for pessimism were 6.8 at T1 and 7.4 at T2.

The family environment was assessed using the Cohesion, Expressiveness, and Conflict subscales of the Family Environment Scale (FES; Moos & Moos 1981). This is a self-report measure that requires respondents to indicate whether each of the 27 items is currently true or false for most family members. Mothers completed this measure. Higher scores indicate less cohesion and expressiveness and more conflict within the relevant subscales. Within the Cohesion subscale, Cronbach's alpha reliability was .74 at T1 and .73 at T2, with the means for the sample being 7.8 and 7.5 at T1 and T2, respectively. For the Expressiveness subscale, Cronbach's alpha reliability was .65 at T1 and .69 at T2, with the means for the sample being 6.8 and 6.2 at T1 and T2, respectively. The Conflict subscale had Cronbach's alpha reliability values of .56 at T1 and .66 at T2, with the means for the sample being 1.9 and 1.5 at T1 and T2, respectively. Note that these alpha coefficients were low for the Conflict subscale at T1, and thus, caution should be exercised in interpreting the results for this subscale.

Maladaptive child behavior was measured through the use of three instruments, each completed by the target youth's father. One measure was the Child Behavior Checklist/6–18 (CBCL/6–18; Achenbach and Rescorla 2001). This is a self-administered informant-report measure in which the parent rates the frequency of 118 problem behaviors exhibited by the child or adolescent on a three point scale ranging from "not true" to "very true or often true." Our data were drawn from the "Withdrawn, Anxious/Depressed," and "Attention Problems" subscales because these symptoms distinguish adolescents with FXS from typically developing adolescents. Internal consistency estimates for the Anxious/Depressed, Withdrawn, and Attention Problems subscales were, respectively .72, .41, and .57 at T1 and .81, .57, and .68 at T2. Note that the alpha coefficients were quite low for the Withdrawn subscale and somewhat low for the Attention Problems subscale; thus, caution should be exercised in interpreting the results for this subscale.

The second measure of maladaptive child behavior was the Autism Behavior Checklist (ABC; Krug et al., 1980), which focused on the presence of behaviors characteristic of autism. This is an informant-report measure containing 57 yes/no items about previous or current child functioning. Higher scores reflect a greater likelihood of autism, with a score of 44 considered the clinical threshold. Cronbach's alpha reliability for this measure was 0.92 at T1 and 0.91 at T2.

The third measure of maladaptive child behavior was the Problem Behavior Scale from the Inventory for Client and Agency Planning (PBS; Bruininks, Hill, Weatherman, & Woodcock, 1986). This self-administered informant-report assessment requires a parent to indicate which of eight problem behaviors is displayed, and how severely, by the target adolescent. Internal consistency estimates for the PBS were .80 at T1 and .87 at T2.

Finally, the Kaufman Brief Intelligence Test (K-BIT; Kaufman and Kaufman 1990) was used to assess maternal IQ. The K-BIT was administered at T1 only.

1.2.3 Characteristics of mothers

Participating mothers ranged in age from 38 to 55 years, with a mean age of 46.6. All of the mothers identified themselves and their children as white/non-Hispanic. Of the 18 mothers,

12 had attained a college degree or beyond. IQ scores on the K-BIT ranged from 96 to 134, with a mean of 111.4. Mothers reported family income on a numerical rating scale in $10,000 increments. The mean level of family income on this scale was an 8.76, which corresponded to an income between $70,001 and $80,000 annually. The mean number of children in addition to the target youth per family was 1.33, with a range from 0 to 4. In addition to the target child with FXS, a mean of 0.61 of the siblings were diagnosed with a disability, with a range from 0 to 2.

The mean age of the children in our sample was 15.68 years, with the youngest being 11.4 and the oldest being 20.7 years of age. Four of the target children were females and 14 were males. The mean ABC score was 40.83 at T1 and 41.50 at T2, which fell just below the suggested clinical cut-off for possible autism of 44 (Dykens & Volkmar, 1987). The mean CBCL total score was 61.22 at T1 and 56.88 at T2. These scores indicate that, on average, the target children with FXS displayed maladaptive behavior at the lower end of the borderline clinical range (T scores of 60 to 63) at T1 (Achenbach & Rescorla, 2001). The mean score from the PBS was −8.67 at T1 and −9.44 at T2, which are at the higher end of the normal range of behavior and approaching “marginally serious” (i.e., a score of −10).

1.3.1 Results

We utilized the various measures of maternal mental health and family environment individually, while synthesizing the data from our three child behavior measures (CBCL, ABC, and PBS) into a single child behavior composite score. The child behavior composite scores were created by first transforming the summary scores from the CBCL, ABC, and PBS into Z-scores. Higher levels of child challenging behavior were indicated with higher scores on the CBCL and ABC but with lower scores on the PBS; therefore, we reverse scored the PBS Z-scores so that higher scores also indicated higher levels of child challenging behavior. The final step in creating the child challenging behavior composite score was averaging these three Z-scores at T1 and T2.

1.3.2 Prediction of Change in Child Challenging Behavior

Our first research question focused on whether maternal mental health status (including symptoms of psychopathology, depression, and Axis I diagnoses) are related to changes in the levels of child challenging behavior from T1 to T2. In particular, the T1 child challenging behavior composite was subtracted from the T2 version of the composite, with the result being that positive scores reflected a worsening of challenging behavior, whereas negative scores reflected an improvement. Three multiple linear regressions were calculated to determine if change in child challenging behavior from T1 to T2 was predicted by each of three measures of maternal mental health status at T1: the SCL-90-R, the SCID, and the CES-D. One maternal mental health measure was included in each regression. Maternal age and maternal IQ were included as control variables in each regression.

None of the regression equations were significant. The model predicting the child challenging behavior composite from the SCL-90-R, maternal age, and maternal IQ yielded an $F(3,14) = 1.54$, $p = .249$, and $r^2 = .25$; the model predicting in the child challenging behavior composite from the SCID and the other two maternal variables yielded an $F(3,14)$

= 1.77, p = .198, and r^2 = .28; and the model predicting the child challenging behavior composite from the CES-D and the other two maternal variables yielded an, $F(3,14) = 1.53$, p = .250, and r^2 = .25.

1.3.3 Prediction of Change in Maternal Mental Health Status

The second research question focused on whether child challenging behavior is related to changes in maternal mental health status from T1 to T2. The SCL-90-R and CES-D were used to index maternal mental health status because they were given at both time points, whereas the SCID was only administered at T1. Two dependent variables were created by subtracting T1 from T2 values for the SCL-90-R and for the CES-D. Again, positive difference scores indicated a worsening of symptoms and negative scores, an improvement. For each of these dependent variables, a multiple regression was conducted in which the child challenging behavior composite for T1 was a predictor along with maternal age and maternal IQ.

The first regression examined the relationship between child challenging behavior at T1 and change in maternal mental health, as measured by the SCL-90-R. This model approached, but did not reach significance, $F(3,14) = 2.42$, p = .110, r^2 = .34. In the regression in which change in maternal CES-D scores was the dependent variable, however, the model was significant, $F(3,14) = 5.52$, p = .010, r^2 =.542. Maternal IQ (β = .63, t = 3.05, p = .009), maternal age (β = -.48, t = −2.28, p = .038), and child behavior (β = -.41, t = −2.25, p = .041) all significantly contributed to the model. Note that the relationship between CES-D change and both child behavior and maternal age was negative; that is, more child challenging behavior and older maternal age at T1 were associated with improvement in maternal depression scores.

1.3.4 Prediction of Change in Family Environment

Our final research question focused on whether maternal mental health status and child challenging behavior predict change in the family environment from T1 to T2. For these analyses, we predicted change from T1 to T2 (i.e. T2 values minus T1 values) in each subscale of the Family Environment Scale (Cohesion, Expressiveness, and Conflict), the Pessimism subscale of the Questionnaire on Resources and Stress, and the two subscales of the PAI separately from the child behavior composite and maternal mental health, with each regression repeated three times, once for each of our three measures of maternal mental health (SCL-90-R, SCID, and CES-D). Thus, we calculated a total of 18 multiple linear regression equations. This large number of analyses requires caution in interpreting the results of any one analysis.

Only three regressions were significant; however, all three involved the same outcome variable; namely, maternal feelings of closeness toward the child on the PAI. First, the regression in which child challenging behavior and the SCL-90-R were used to predict change in maternal feelings of closeness towards the child was significant, $F(2,15) = 5.63$, p = .015, r^2 = .43. Within this regression equation, child challenging behavior was a significant predictor (β = .71, t = 3.27, p = .005). Second, the regression in which child challenging behavior and the SCID were used to predict change in maternal feelings of

closeness towards the child was significant, $F(2,15) = 5.23$, $p = .019$, $r^2 = .41$. Again, child challenging behavior was a significant predictor ($\beta = .63$, $t = 3.19$, $p = .006$). Finally, the regression in which child challenging behavior and the CES-D were used to predict change in maternal feelings of closeness towards the child was significant, $F(2,15) = 5.13$, $p = .020$, $r^2 = .406$. Child challenging behavior significantly contributed to the regression equation ($\beta = .65$, $t = 3.16$, $p = .006$). These three findings collectively suggest that change in maternal feelings of closeness towards her child can be predicted from previously measured child challenging behavior, when controlling for maternal mental health status. Note, however, that the relationship was positive, with higher rates of T1 child challenging behavior associated with an increase in maternal feelings of closeness from T1 to T2.

1.4.1 Discussion

The purpose of this study was to investigate three questions regarding the relationships among maternal mental health status, child challenging behavior, and the family environment for families of adolescents with FXS. To provide insight into the direction of influence among the variables, we conducted a short-term longitudinal study with a small sample of adolescents with FXS and their biological mothers, who carried either the premutation or in a few cases, had mosaic carrier status (i.e., carried both premutation and full mutation cells).

Our first research question was whether maternal mental health status at T1 could predict change in child challenging behavior from T1 to T2; over an approximately one-year interval. Previous studies have established concurrent relationships between maternal depression and problematic child behavior (NICHD 1999), and even some evidence that maternal psychological status can affect child challenging behavior over time (e.g., Orsmond, Seltzer, Krauss, & Hong 2003). In the present study, we found no evidence that maternal mental health status predicted change in child challenging behavior for families affected by FXS. Moreover, the same null finding emerged whether we used self-report or clinician-conducted psychiatric interviews to assess maternal mental health and whether we considered broad summary measures or only symptoms of depression. It may be that the challenging behaviors of adolescents with FXS are so well-entrenched that they are resistant to the environmental variations correlated with maternal mental health or are controlled by regulatory mechanisms “internal” to the adolescent with FXS rather than by the environment. It may also be that in the short-term, mothers are able to prevent their own stress and mental health challenges from affecting their interactions with the adolescent with FXS. Deciding among these possibilities awaits further research.

The second research question focused on whether child challenging behavior at T1 could predict change in maternal mental health status from T1 to T2. We found no evidence that child challenging behavior predicted change in maternal mental health assessed with the SCL-90-R, a self-report measure of a range of mental health symptoms and conditions, including anxiety and obsessive-compulsive disorders. We did find, however, that change in maternal symptoms of depression, as measured by the CES-D, was predicted from child challenging behavior at T1, even after controlling for the contributions of maternal age and

IQ. Thus, there appears to be specificity in the relationship between child challenging behavior and maternal mental health status.

The finding that maternal depressive symptom change can be predicted from previous child challenging behavior supports the results of Bailey, Sideris, Roberts, and Hatton (2008) and Abbeduto et al. (2004), in which the child's behavioral symptoms were the strongest predictor of maternal depressive symptoms. The current finding also reinforces the results of Wheeler et al. (2010) in which child challenging behavior rates were highly correlated with both parenting stress and maternal depression. The present finding, however, is the first to focus on longitudinal rather than concurrent relationships and thus, the first to provide evidence suggestive of a direction of impact between these variables for families affected by FXS.

Contrary to expectations, however, we found that higher rates of child challenging behavior were associated with a decrease in maternal symptoms of depression (on the CES-D) over time. One possible explanation for this finding is that high rates of adolescent challenging behavior may bring additional resources and support to the mother, either in the form of additional help from the adolescent's father, other family members, or even professionals. This additional support may serve to lower maternal burden and stress, thereby leading to improved maternal mental health. Stated somewhat differently, relatively low levels of challenging behavior may represent a chronic stressor that mothers are left to deal with on their own and with considerable cost to maternal mental health. If verified empirically, this hypothesis would suggest that the threshold for determining when mothers of adolescents with FXS could benefit from therapeutic intervention may need to be set lower than it is currently.

Our final research question focused on whether maternal mental health status and child behavior predict change in the emotional climate of the family, particularly in terms of levels of family cohesion, expressiveness and conflict, maternal pessimism regarding the adolescent's future, and closeness within the mother/child relationship. Here too, our findings were contrary to expectations. We did find that changes in maternal feelings of closeness toward her adolescent child were predicted by adolescent challenging behavior. Moreover, this finding was consistent across three analyses that included three different measures of maternal mental health, including symptoms of psychopathology (SCL-90-R), DSM-IV diagnoses (SCID), and symptoms of depression (CES-D). Unexpectedly, however, higher adolescent challenging behavior scores at T1 were associated with increased feelings of closeness toward the adolescent on the part of mothers. The PAI items that measure maternal feelings of closeness include "understanding your child, trusting your child, feeling that you are fair towards your child, respecting your child, and having affection towards your child." These qualities of the mother-child relationship may be enhanced by behavioral problems because the mother is spending more time with the adolescent, more of her attention is devoted to the adolescent, and she may feel a greater need to serve as the adolescent's advocate and protector. In short, the mother may view the adolescent's challenging behaviors as isolating the adolescent from others and making him or her more dependent on the mother. It is also possible that mothers of adolescents with more challenging behavior may be pulled to answer questions about their relationship with their

adolescent in a manner that more strongly reflects the positive aspects of their relationship (i.e., a positive response bias). Deciding between these alternatives will require additional research.

1.5.1 Conclusions

In summary, our findings suggest that mothers of adolescents with FXS are highly resilient in the face of many challenges. Indeed, we speculate that the adolescent's challenging behaviors may trigger a marshaling of resources through other people or perhaps even maternal psychological resources, with the result being enhanced feelings of closeness toward the adolescent on the mother's part and improvement in maternal symptoms of depression over time. It would be helpful for future studies to examine the factors that contribute to this resiliency in order to better identify sources of family strength and develop specific treatment objectives. At the same time, it would be important to determine whether what we have identified is simply a short-term adaptation. Can maternal well being continue to be positive in the face of the persistent high rates of challenging behaviors presented by their adolescent sons and daughters?

Our study has several strengths, including a) the longitudinal design necessary to examine ongoing relationships among these variables; b) the use of fathers as informants about adolescent challenging behavior to avoid the problem of a shared informant across constructs; and c) the use of multiple methods for assessing maternal mental health. Although the use of father report on child behavior addresses possible mono-reporter bias, this approach also has its limitations. In particular, the biological mother was the primary careprovider for the individual with FXS in all participating families. This raises the possibility that fathers in our study may have inaccurate or biased representation of the child's functioning based on their more limited interactions and experience. In order to address this, it would be prudent to include both maternal and paternal reports of child behavior and family functioning in future studies. The addition of this data would allow researchers to ensure consistency across parent reports of child behavior and thereby account for this potential confound.

Other limitations also must be acknowledged, including the small sample size, the computation of a large number of statistical tests, and the relatively brief interval of the longitudinal component. Moreover, the small sample size required us to test only "pieces" of a more comprehensive model and this may have led us to miss more complex relationships and variables that could help explain the relationships observed, particularly those relationships that were contrary to expectations. In addition, the limited number of participants resulted in a sample that is unrepresentative of the larger population of families of children with FXS. Indeed, our maternal sample was well-educated, had obtained above average IQ scores, and the child sample was exclusively White. Moreover, all participating families were presumably functioning well enough to travel, sometimes at great distances, to participate in the study. Therefore, the generalizability of these results is limited.

Finally, some of the informant report measures yielded low estimates of internal consistency for some subscales, meaning that they might not be the most appropriate measures for FXS,

although it is worth noting that few FXS-specific measures exist. Nevertheless, the results suggest a new, more complex understanding, of the ways in which mothers deal with the stress associated with raising a son or daughter with special needs. Clearly, replication and further examination of constructs reflecting resilience and adaptation are warranted.

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Highlights

We examined bidirectional influences between maternal mental health status, maternal stress, family environment and child challenging behavior in families with a son with fragile X syndrome.

A short-term longitudinal design was used, with two-parent families that included the biological mother of the youth with fragile X syndrome. Maternal self-report measures were used to assess maternal mental health, maternal stress, and family environment, whereas fathers reported on child challenging behavior.

Findings indicated that maternal mental health status was not significantly related to subsequent changes in levels of child challenging behavior.

Child challenging behavior was related to subsequent improvement in maternal depression over time, and a heightened level of child challenging behavior was related to increased feelings of maternal closeness toward the child over time. These positive effects of child challenging behavior may be short term positive adaptations resulting from a marshaling of new resources by mothers to deal with child change.