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Change in Maladaptive Behavior Affects Intergenerational Relationships in FXS

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Change in Maladaptive Behavior Affects Intergenerational Relationships in FXS

Abstract

This study investigated the bidirectional effects of change in maladaptive behaviors among adolescents and adults with FXS and change in their intergenerational family relationships over a 7.5-year period. Indicators of the intergenerational family relationship between premutation carrier mothers and their adolescent or adult son/daughter with FXS included a measure of the quality of the relationship, as well as descriptions provided by mothers of their relationship with their son/daughter (positive remarks, critical remarks). Maladaptive behaviors decreased, maternal positive remarks increased, and maternal critical remarks and relationship quality remained stable over time. Bidirectional effects of change were observed in predicting maladaptive behaviors and maternal positive remarks, although maladaptive behaviors more strongly predicted positive remarks than the reciprocal association. This research suggests prioritizing maladaptive behaviors in the context of family interventions.

Introduction

Fragile X syndrome (FXS) is the leading inherited cause of intellectual disability and occurs in approximately one in 3,600 to 4,000 males and one in 4,000 to 6,000 females (Crawford et al., 2001; Kooy et al., 2000; Turner et al., 1996). FXS results from over 200 CGG repeats on the *FMR1* gene on the X chromosome (Verkerk et al., 1991). Due to its X-linked nature, males with FXS are generally more seriously affected than females. Phenotypic features associated with FXS often include intellectual disability, increased symptoms of autism spectrum disorder (ASD), heightened maladaptive behaviors, hyperarousal, and language deficits (Abbeduto et al., 2019; Finestack et al., 2009; Hall et al., 2009; Hardiman & McGill, 2018; Usher et al., 2020). FXS is a lifelong disorder and deficits in these areas extend into adulthood, making the study of individuals with FXS beyond the childhood years critical. Recent work has begun to examine family-level factors (e.g., maternal descriptions of their affective relationship with their son/daughter with FXS and their perception of the quality of the relationship) that may support improvement in functioning in FXS beyond the early years (Baker et al., 2012; Fielding-Gebhardt et al., 2020; Greenberg et al., 2012; Smith et al., 2016). The present study utilized a longitudinal design to investigate the reciprocal effects of change in son/daughter maladaptive behaviors, maternal descriptions of their affective relationship (maternal positive and critical remarks), and relationship quality between the mother and son/daughter with FXS over an extended period of time. The adolescents and adults with FXS were ages 12 to 40 years at Time 1.

The transactional model of development emphasizes the cumulative and dynamic effects that parents and their son/daughter have on one another (Sameroff & MacKenzie, 2003). To date, a substantial body of research exists in typical development and other neurodevelopmental

disabilities showing the bidirectional nature of parent and child factors (Barnett et al., 2012; Greenberg et al., 2006; Newton et al., 2014; Orsmond et al., 2003). While much of the research using this theoretical model has focused on interactions early in childhood and over relatively brief periods of time, research suggests that this complex interplay extends into adolescence and adulthood (Greenberg et al., 2012; Hauser et al., 2014; Smith et al., 2016). The transactional model is particularly important to study in FXS due to the heritable and genetic nature of the disorder which often results in multiple family members being impacted by *FMRI* mutations. Specifically, the parenting experience of mothers of **individuals** with FXS is unique since most of these mothers are *FMRI* premutation carriers (i.e., 55-200 CGG repeats). Premutation carriers are at an increased risk for a range of mental and physical health problems, such as depression, anxiety, and menstrual-related symptoms (Bourgeois et al., 2011; Movaghar et al., 2019; Roberts et al., 2009; Wheeler et al., 2014), although not all premutation carriers are equally affected (Klusek et al., 2020; Roberts et al., 2016).

The transactional model also remains relevant throughout the lifespan in FXS because many deficits associated with the disorder (e.g., language difficulties, heightened anxiety) extend into adolescence and adulthood (Abbeduto et al., 2019; Ezell et al., 2019; Roberts et al., 2009). Children, adolescents, and adults with FXS present with elevated rates of clinical-level maladaptive behaviors (for a review, see Hardiman & McGill, 2018), although these behaviors may decrease over time (Fielding-Gebhardt et al., 2020; Usher et al., 2020). High levels of maladaptive behaviors can interact with genetic risk to exacerbate symptoms of mood disorders in mothers (Bailey et al., 2008; Baker et al., 2012; Fielding-Gebhardt et al., 2020; Roberts et al., 2016). A study of FXS over nearly seven years found that despite stability in maternal mental health and relationship quality **between the mother and son/daughter**, within-child change and

between-child variability in maladaptive behaviors in combination with between-mother variability in mental health (i.e., symptoms of anxiety and depression) predicted relationship quality (Fielding-Gebhardt et al., 2020). Unexpectedly, Hauser et al. (2014) found that in a sample of mothers and their **sons/daughters** with FXS ages 11 to 20 years, more severe maladaptive behaviors at Time 1 predicted improvements in relationship quality (i.e., maternal closeness) over one year, but this is the only study to report such an association.

Families of **individuals** with FXS experience increased levels of parenting stress and the prolonged presence of maladaptive behaviors (Bailey et al., 2008; Hardiman & McGill, 2018), which might challenge the relationship **between the mother and her son/daughter**. Specifically, previous studies in FXS have linked maternal criticism in speech samples to maladaptive behaviors both cross-sectionally and longitudinally (Greenberg et al., 2012; Smith et al., 2016), although findings from Smith et al. (2016) found no effect of *change* in maternal criticism on later maladaptive behaviors three years later. Greenberg et al. (2012) looked cross-sectionally and found a negative association between maternal positive remarks in the speech samples and maladaptive behaviors in children and adults with FXS. These findings suggest that maternal affect toward her **son/daughter** may shape the **son/daughter's** behaviors in important ways, which is in line with previous research indicating that parenting style impacts child maladaptive behaviors (Rinaldi & Howe, 2012). However, it is not yet clear how the associations between maternal descriptions of the relationship, **the relationship quality between the mother and son/daughter**, and maladaptive behaviors unfold over an extended period of time.

Purpose of the Present Study

Previous research has largely focused on how earlier time points predict later outcomes, which is important work, yet to the best of our knowledge, no past study has taken into account

the role of change above and beyond the initial level of the variables into later adolescence and adulthood. Using well-controlled statistical models, the current study investigated bidirectional effects, and focused on the role of change above and beyond initial level, as well as the relative strength of change in maladaptive behaviors, maternal **positive and critical remarks**, and relationship quality **between the mother and son/daughter** in predicting outcomes 7.5 years later. The direction and strength of effects may not mirror findings from studies of the general population. Describing these relationships within the FXS population can provide insight into ways to support two clinically relevant populations – **individuals** with FXS and their mothers with the *FMRI* premutation. This work can provide insight into interdependent intergenerational family relationships over an extended period of time, potentially revealing direction of effects and thus identifying intervention targets for family-based interventions. Therefore, we asked the following questions:

1. How do maternal positive remarks, maternal critical remarks, and relationship quality change over 7.5 years in mothers of adolescents and adults with FXS?
2. How do maladaptive behaviors change over 7.5 years in adolescents and adults with FXS?
3. To what extent does change in maternal positive remarks, maternal critical remarks, and relationship quality over 7.5 years predict maladaptive behaviors? To what extent does change in maladaptive behaviors over this time period predict maternal positive remarks, maternal critical remarks, and relationship quality? Of these potentially bidirectional relationships, which is the relatively stronger direction of effects?

Method

Procedure

Mothers of adolescents and adults with FXS participated in a larger ongoing longitudinal study of family adaptation to fragile X syndrome (REMOVED FOR PEER REVIEW). Study procedures were approved by [REMOVED FOR PEER REVIEW] Institutional Review Board. Families were recruited through university disability research registries, FXS foundations, service agencies, and clinics. For the current study, inclusionary criteria included the following: the mother was the biological parent of the son or daughter with FXS, the mother was a premutation carrier of the *FMR1* gene, the son/daughter had the *FMR1* full mutation, the son/daughter lived with the mother continuously across the four study time points (i.e., from Time 1 to Time 4), and the mother participated in the study at each of the four time points. These inclusionary criteria resulted in a sample size of 78 mother-adolescent/adult child dyads. We only included dyads where the son/daughter was continuously co-residing with the mother in order to ensure daily exposure and frequent interactions between the mother and her son/daughter throughout study participation.

We completed an attrition analysis to examine differences between those who originally met criteria at Time 1 but dropped out or had incomplete data at subsequent time points. The attrition cases ($n = 36$) did not significantly differ from the current sample on maternal education, $p = .524$, marital status, $p = .065$, son/daughter age, $p = .826$, maternal age, $p = .282$, or income, $p = .137$.

Data were obtained at all four time points. All mothers provided written informed consent prior to participating. At each time point, mothers were interviewed for approximately one hour via telephone. Mothers also completed several parent-report measures on their son/daughter as well as self-report measures. When a mother had more than one son/daughter with FXS, she was asked to report on the son/daughter living in the home. If multiple children with FXS lived in the

home, the mother was asked to report on the **son/daughter** she viewed as most severely affected. For the current study, we primarily utilized data from Time 1 and Time 4, which occurred an average of 7.5 years apart. Continuous coresidence of the **son/daughter** with FXS and the mother was confirmed at the intervening time points as well as at Times 1 and 4.

Participants

Ages for the adolescents and adults with FXS ranged from 12 to 40 years ($M = 19.72$, $SD = 6.59$) at Time 1 and 19 to 47 years ($M = 27.00$, $SD = 6.59$) at Time 4. The majority were sons ($n = 67$, 85.90%), had an intellectual disability ($n = 74$, 94.87%), and were white ($n = 74$, 94.87%). All mothers provided genetic documentation of their **son/daughter's** *FMRI* full mutation status.

Maternal ages ranged from 36 to 67 years ($M = 49.66$, $SD = 7.11$) at Time 1 and 44 to 75 years ($M = 57.19$, $SD = 7.08$) at Time 4. All 78 mothers were premutation carriers of the *FMRI* gene based on genetic testing. The majority of mothers were married at Time 1 ($n = 67$, 85.90%) as well as Time 4 ($n = 67$, 85.90%). Ten mothers had a high school degree, 47 had some college or a bachelor's degree, and 21 had a post-bachelors or graduate degree. The majority of mothers were white ($n = 75$, 96.15%). Median household income was between \$90,000 and \$99,999 at Time 1 and Time 4, although a substantial range was represented (less than \$9,999 to greater than \$160,000).

Measures

Maternal description of her affective relationship with her son/daughter. All mothers participated in a Five Minute Speech Sample (FMSS; Magaña et al., 1986) during the phone interview at each time point. The FMSS consists of a specific, standardized prompt that instructs the caregiver to describe his or her relationship with the person with the disability and his or her

thoughts and feelings about that individual for five minutes without interruption. This measure was recorded, transcribed, and further coded to measure the parent-child relationship. We utilized previously developed coding procedures by Magaña et al. (1986); coding included the identification of both positive and critical remarks. Using the FMSS to measure maternal descriptions of the **affective relationship with** one's child is consistent with previous work in families of **individuals** with neurodevelopmental disorders (Beck et al., 2004; Greenberg et al., 2012; Hastings et al., 2006; Smith et al., 2008; Woodman et al., 2015).

Positive remarks. When coding the FMSS, a rater with over 30 years of experience who was **not informed of the son/daughter's** diagnosis or the purpose of the study identified the total number of positive remarks that each mother made about her son or daughter. A positive remark was defined as a positive statement or praise of the **son/daughter's** behaviors or characteristics. The raw number of positive remarks ranged from zero to 16 at Time 1 and zero to 25 at Time 4.

Critical remarks. The same rater identified the total number of critical remarks that each mother made about her son or daughter throughout the FMSS. A critical remark was defined as a negative statement about the **son/daughter's** behaviors or characteristics. The raw number of critical remarks ranged from zero to three at Time 1 and zero to two at Time 4.

Reliability. Interrater reliability based on absolute agreement and single-measure values for intraclass correlation coefficients between the primary rater and another trained rater on FMSS ratings was calculated based on seven files (i.e., 4.5% of the sample). ICCs indicated excellent agreement between raters for maternal positive remarks, $ICC = .89$, and maternal critical remarks, $ICC = 1.00$ (Hallgren, 2012).

Mother-child relationship quality. Each mother completed the Bengtson Positive Affect Index at Time 1 and Time 4 (Bengtson & Schrader, 1982). The Positive Affect Index is a

10-item parent-report measure used to index relationship quality **between the mother and son/daughter** and has been used in previous work within FXS and neurodevelopmental disabilities more broadly (Esbensen et al., 2013; Fielding-Gebhardt et al., 2020; Hartley et al., 2011; Smith et al., 2008). Feelings of trust, affection, understanding, fairness, and respect are rated twice – in terms of the mother’s feelings of closeness toward her **son/daughter** and also the extent to which each mother perceives that her **son/daughter** displays each feeling toward her. Each item is rated on a six-point scale with higher numbers indicating better relationship quality. Cronbach’s alpha was .78 and .87 at Time 1 and Time 4, respectively.

Maladaptive behaviors. We utilized the Problem Behavior subscale of the Scales of Independent Behavior – Revised (SIB-R; Bruininks et al., 1996) to measure behavior problems in the son/daughter with FXS. The Problem Behavior subscale assesses eight types of problem behaviors, including (1) hurtful to self, (2) unusual or repetitive, (3) withdrawn or inattentive, (4) destructive to property, (5) hurtful to others, (6) disruptive, (7) socially offensive, and (8) uncooperative. Mothers were asked whether their **son/daughter** displayed episodes of each type of problem behavior in the last six months and if so, to rate the frequency (1-5, where 1=less than once a month and 5=one or more times an hour) and severity (1-5, where 1=not serious and 5=extremely serious) of the behavior. Based on standardized algorithms (Bruininks et al., 1996), three subscales representing three distinct behavioral domains were computed at Time 1 and Time 4: internalized maladaptive behaviors (withdrawn or inattentive, hurtful to self, unusual or repetitive behaviors), externalized maladaptive behaviors (hurtful to others, destructive to property, disruptive behaviors), and asocial maladaptive behaviors (socially offensive, uncooperative behaviors). The SIB-R adjusts for age and higher scores indicate more frequent and severe behavior problems. Values range from 90 to 150, with scores at or above 110

considered above the clinical cutoff for significant behavior problems. Reliability and validity were previously established (Bruininks et al., 1996). One family had missing data from SIB-R items contributing to the Time 4 externalized maladaptive subscale. Therefore, analyses involving that measure at Time 4 included 77 families.

Data Analysis Plan

Prior to analyses, model assumptions were examined; regarding normality, three of 18 key variables (Time 1 maternal critical remarks, Time 4 maternal critical remarks, and Time 4 externalized maladaptive behaviors) were <0.5 outside the acceptable range in skewness while the remaining variables were within the acceptable range for skewness; all of the key variables were within the acceptable range for kurtosis. Because parametric tests are robust to violations of normality and have more statistical power than nonparametric tests, we elected to conduct parametric statistics throughout (Rasch & Guiard, 2004).

Using G*Power (Faul et al., 2007), power analyses based on 78 mother-adolescent/adult child dyads revealed that power to detect a large effect in paired samples *t*-tests and multiple regression models with four predictors was .99 and power to detect a medium effect ranged from .76 - .78.

Research question 1. We used two approaches to examine change in maternal positive remarks, maternal critical remarks, and relationship quality between the mother and her son/daughter with FXS. To examine group-level change from Time 1 to Time 4, we used paired samples *t*-tests. To examine individual-level change, we calculated standardized mean differences (*d*) between Time 1 and Time 4 maternal positive remarks, maternal critical remarks, and relationship quality using the Time 1 standard deviations as the standardizers. This approach is recommended for dependent samples (Kline, 2004) and represents differences between Time 1

and Time 4 means as expressed in standard deviation units. A standardized mean difference of .5 indicates that the Time 4 value was half a standard deviation above the Time 1 value. Utilizing standardized mean differences separately for positive remarks, critical remarks, and relationship quality, mothers were subsequently categorized into three categories: improved, no change, or worsened. The “no change” group constituted mothers whose Time 4 scores were within half the Time 1 standard deviation above or below the Time 1 score (i.e., $-.50 < d < .50$). The mothers whose Time 4 scores changed more than half a standard deviation were classified as “improved” or “worsened.” Specifically, for maternal positive remarks and relationship quality, $d \geq .50$ resulted in categorization into the “improved” group and $d \leq -.50$, resulted in categorization into the “worsened” group. For maternal critical remarks, $d \geq .50$ resulted in categorization into the “worsened” group and $d \leq -.50$, resulted in categorization into the “improved” group since an increase in maternal critical remarks would be interpreted as worsening. Rationale for defining groups based on half a standard deviation is based on work indicating that half a standard deviation of change is clinically meaningful in behavioral research (Norman et al., 2003). The half standard deviation increment is also consistent with a medium effect as defined by Cohen (1988). This approach is also consistent with previous work in neurodevelopmental disabilities (Shattuck et al., 2007; Woodman et al., 2015).

Research question 2. The same statistical approaches described above for research question 1 were utilized to answer the second research question (i.e., paired samples *t*-tests and group categorization based on standardized mean differences). We also reported the number and percentage of individuals with FXS that exceeded cutoff scores on each maladaptive behavior subscale at Time 1 and Time 4 in order to understand the percentage of **individuals** within the clinical range in each domain.

Research question 3. To examine the bidirectional effects between each of the three maladaptive behavior subscales and maternal positive remarks, critical remarks, and relationship quality, we constructed multiple regression. *The multiple regressions were estimated using seemingly unrelated regressions (SUR) models (see additional details below; Zellner, 2006).* Son/daughter chronological age was controlled in all models due to past research indicating age-related differences in *maternal positive and negative remarks* and *son/daughter* behaviors (Greenberg et al., 2012; Usher et al., 2020). *Son/daughter* sex was not included as a covariate since preliminary analyses did not show an effect of sex on the key study variables. The multiple regression models were structured as follows: first, Time 1 levels and changes in the level of *maternal positive or negative remarks* or relationship quality between Time 1 and Time 4 (computed by subtracting Time 1 from Time 4) were used to predict Time 4 *son/daughter* maladaptive behaviors, controlling for Time 1 maladaptive behaviors and *son/daughter* age. Next, the models were reversed to predict maternal variables from *son/daughter* variables. Specifically, Time 1 levels and changes in the level of *son/daughter* maladaptive behaviors between Time 1 and Time 4 were used to predict *maternal positive or negative remarks* or relationship quality, controlling for Time 1 *maternal positive or negative remarks* or relationship quality and *son/daughter* age. By including the Time 1 value of the independent variable as well as the change score, this approach allowed for interpretation of the effect of change above and beyond the initial position of the independent variable. In addition, inclusion of the Time 1 value of the dependent variable accounted for stability effects within each mother and her *son/daughter*.

The pairs of multiple regressions described above were estimated simultaneously using SUR models (Zellner, 2006), which are one type of structural equation models used to examine

bidirectional effects. For example, to evaluate the association between internalized maladaptive behaviors and maternal positive remarks, one SUR model included the multiple regression model predicting **son/daughter** internalized maladaptive behaviors from maternal positive remarks and the multiple regression model predicting maternal positive remarks from **son/daughter** internalized maladaptive behaviors. Similarly, we estimated SUR models to test bidirectional effects between maternal positive remarks and the two other maladaptive behavior domains (i.e., externalized and asocial maladaptive behaviors), maternal critical remarks and each domain of maladaptive behaviors, and relationship quality **between the mother and her son/daughter** and the three domains of maladaptive behaviors (i.e., nine total SUR models; see Tables 3, 4, & 5).

SUR models are particularly useful when examining bidirectional effects because they allow for direct statistical testing of the relative strength of effects across regression models to determine if one directional effect is statistically greater than the other (Zellner, 2006).

Therefore, when change scores significantly predicted Time 4 outcomes, we used model restrictions within the SUR models to test whether change in maladaptive behaviors predicted Time 4 maternal **positive or critical remarks** or relationship quality to the same degree that change in **positive or critical remarks** or relationship quality predicted Time 4 maladaptive behaviors. Through this approach, we determined the relative strength of the bidirectional effects (i.e., whether change in **son/daughter** behaviors more strongly predicted **positive or critical remarks** or relationship quality, or vice versa). For additional details, see Zellner (2006).

Results

Research Question 1: Change in Maternal Positive Remarks, Maternal Critical Remarks, and Relationship Quality Over 7.5 Years

When examining group-level change, we found that maternal positive remarks significantly increased from Time 1 to Time 4 (see Table 1). There were no significant differences between Time 1 and Time 4 maternal critical remarks or relationship quality ratings. When examining individual-level change, we found that more than half of the sample (60.26%) improved in the number of maternal positive remarks, approximately a quarter of the sample (24.36%) showed no change, and an even smaller percentage (15.38%) worsened. In terms of the number of maternal critical remarks, the majority of the sample (80.77%) showed no change; a small percentage improved (11.54%) or worsened (7.69%). In terms of relationship quality **between the mother and her son/daughter**, slightly less than half of the sample (41.03%) showed no change, and slightly over a quarter of the sample improved (29.49%) or worsened (29.49%).

Research Question 2: Change in **Son/Daughter Maladaptive Behaviors Over 7.5 Years**

When examining group-level change, we found that internalized maladaptive behaviors, externalized maladaptive behaviors, and asocial maladaptive behaviors all decreased significantly from Time 1 to Time 4 (see Table 1). Regarding clinically significant internalized maladaptive behaviors, we found that 52.56% ($n = 41$) and 34.62% ($n = 27$) of individuals with FXS met the cutoff at Time 1 and Time 4, respectively. Regarding externalized maladaptive behaviors, 19.48% ($n = 15$) and 12.99% ($n = 10$) of individuals with FXS met this cutoff at Time 1 and Time 4, respectively. For asocial maladaptive behaviors, 42.31% ($n = 33$) and 32.05% ($n = 25$) of individuals with FXS met this cutoff at Time 1 and Time 4, respectively.

We examined individual-level change for each maladaptive behavior subscale (see Table 2). With regard to internalized maladaptive behaviors, approximately half of the sample (48.72%) showed improvement from Time 1 to Time 4, slightly more than a third of the sample (35.90%) showed no change, and a relatively smaller percentage (15.38%) worsened. In terms of

externalized maladaptive behaviors, the majority of the sample (57.14%) showed no change, slightly more than a quarter of the sample (28.57%) showed improvement, and a relatively small percentage of participants (14.29%) worsened. In terms of asocial maladaptive behaviors, approximately half of the sample (47.44%) showed no change, more than a third of the sample (37.18%) showed improvement, and a relatively small percentage of participants (15.38%) worsened.

Research Question 3: Bidirectional Effects between Maternal **Positive and Critical Remarks, Relationship Quality, and **Son/Daughter Maladaptive Behaviors****

Maternal positive remarks and maladaptive behaviors. See Table 3 for unstandardized coefficients (β), standard errors, and p -values associated with the SUR models.

Internalized maladaptive behaviors. When estimating the SUR model, the multiple regression model predicting Time 4 internalized maladaptive behaviors accounted for 47.80% of the variance, $F(4, 73) = 30.02, p < .001$. Greater increases in maternal positive remarks from Time 1 to Time 4 was associated with less severe Time 4 internalized maladaptive behaviors. Reciprocally, the multiple regression model predicting Time 4 maternal positive remarks accounted for 17.71% of the variance, $F(4, 73) = 16.06, p < .001$. Greater decreases in internalized maladaptive behaviors from Time 1 to Time 4 were associated with more Time 4 maternal positive remarks. Results of model restrictions indicated that change in internalized maladaptive behaviors over time had a significantly stronger impact on Time 4 maternal positive remarks than change in maternal positive remarks had on Time 4 internalized maladaptive behaviors, $F(1, 146) = 27.63, p < .001$.

Externalized maladaptive behaviors. When estimating the SUR model, the multiple regression model predicting Time 4 externalized maladaptive behaviors accounted for 33.93% of

the variance, $F(4, 72) = 12.77, p < .001$. Greater increases in maternal positive remarks from Time 1 to Time 4 was associated with less severe Time 4 externalized maladaptive behaviors. Reciprocally, the multiple regression model predicting Time 4 maternal positive remarks accounted for 9.63% of the variance, $F(4, 72) = 4.74, p = .001$. Greater decreases in externalized maladaptive behaviors from Time 1 to Time 4 were associated with more Time 4 maternal positive remarks. Results of model restrictions indicated that change in externalized maladaptive behaviors over time had a significantly stronger impact on Time 4 maternal positive remarks than change in maternal positive remarks had on Time 4 externalized maladaptive behaviors, $F(1, 144) = 3.96, p = .049$.

Asocial maladaptive behaviors. When estimating the SUR model, the multiple regression model predicting Time 4 asocial maladaptive behaviors accounted for 23.01% of the variance, $F(4, 73) = 9.45, p < .001$. Greater increases in maternal positive remarks from Time 1 to Time 4 was associated with less severe Time 4 asocial maladaptive behaviors. Reciprocally, the multiple regression model predicting Time 4 maternal positive remarks accounted for 9.32% of the variance, $F(4, 73) = 5.61, p < .001$. Greater decreases in asocial maladaptive behaviors from Time 1 to Time 4 were associated with more Time 4 maternal positive remarks. Results of model restrictions indicated that change in asocial maladaptive behaviors over time had a significantly stronger impact on Time 4 maternal positive remarks than change in maternal positive remarks had on Time 4 asocial maladaptive behaviors, $F(1, 146) = 8.73, p = .004$.

Maternal critical remarks and maladaptive behaviors. See Table 4 for unstandardized coefficients (β), standard errors, and p -values associated with the SUR models.

Internalized maladaptive behaviors. When estimating the SUR model, the multiple regression model predicting Time 4 internalized maladaptive behaviors accounted for 42.42% of

the variance, $F(4, 73) = 14.98, p < .001$. The multiple regression model predicting Time 4 maternal critical remarks accounted for 21.36% of the variance, $F(4, 73) = 5.86, p < .001$. No bidirectional effects were observed between maternal critical remarks and internalized maladaptive behaviors.

Externalized maladaptive behaviors. When estimating the SUR model, the multiple regression model predicting Time 4 externalized maladaptive behaviors accounted for 30.87% of the variance, $F(4, 72) = 8.60, p < .001$. The multiple regression model predicting Time 4 maternal positive remarks accounted for 22.72% of the variance, $F(4, 72) = 5.67, p < .001$. No bidirectional effects were observed between maternal critical remarks and externalized maladaptive behaviors.

Asocial maladaptive behaviors. When estimating the SUR model, the multiple regression model predicting Time 4 asocial maladaptive behaviors accounted for 22.11% of the variance, $F(4, 73) = 6.31, p < .001$. The multiple regression model predicting Time 4 maternal critical remarks accounted for 21.54% of the variance, $F(4, 73) = 6.13, p < .001$. No bidirectional effects were observed between maternal critical remarks and asocial maladaptive behaviors.

Relationship quality and maladaptive behaviors. See Table 5 for unstandardized coefficients (β), standard errors, and p -values associated with the SUR models.

Internalized maladaptive behaviors. When estimating the SUR model, the multiple regression model predicting Time 4 internalized maladaptive behaviors accounted for 42.09% of the variance, $F(4, 73) = 15.08, p < .001$. The multiple regression model predicting Time 4 relationship quality accounted for 36.47% of the variance, $F(4, 73) = 12.07, p < .001$. No bidirectional effects were observed between relationship quality and internalized maladaptive behaviors.

Externalized maladaptive behaviors. When estimating the SUR model, the multiple regression model predicting Time 4 externalized maladaptive behaviors accounted for 31.02% of the variance, $F(4, 72) = 9.08, p < .001$. The multiple regression model predicting Time 4 relationship quality accounted for 35.88% of the variance, $F(4, 72) = 11.20, p < .001$. No bidirectional effects were observed between relationship quality and externalized maladaptive behaviors.

Asocial maladaptive behaviors. When estimating the SUR model for asocial maladaptive behaviors, the multiple regression model predicting Time 4 asocial maladaptive behaviors accounted for 22.96% of the variance, $F(4, 73) = 9.82, p < .001$. Greater increases in relationship quality from Time 1 to Time 4 was associated with less severe Time 4 asocial maladaptive behaviors. Reciprocally, the multiple regression model predicting Time 4 relationship quality accounted for 35.58% of the variance, $F(4, 73) = 14.99, p < .001$. Greater decreases in asocial maladaptive behaviors from Time 1 to Time 4 were associated with higher Time 4 relationship quality. Results of model restrictions indicated that change in asocial maladaptive behaviors over time had a significantly stronger impact on Time 4 relationship quality than change in relationship quality had on Time 4 asocial maladaptive behaviors, $F(1, 146) = 6.77, p = .010$.

Discussion

This study extends previous findings in typical development and neurodevelopmental disabilities by identifying bidirectional effects over 7.5 years between maladaptive behaviors of the son/daughter with FXS and both maternal positivity and relationship quality, but not maternal critical remarks. This study also provides novel evidence in support of the transactional model within FXS by delineating the specific aspects of maternal **relationship descriptions** that are associated with maladaptive behaviors in adolescence and adulthood, and the relative strength of

the bidirectional effects. Further, our study found evidence supporting the cumulative effects posited in the transactional model; specifically, we identified long-term bidirectional effects of maladaptive behaviors, maternal positivity, and relationship quality over an extended period of time and well into adolescence and adulthood. Although the effects were bidirectional, we found a stronger impact of change in maladaptive behaviors on maternal positive remarks and relationship quality than the reverse. **These findings are consistent with developmental work showing that maladaptive behaviors decrease over time (Usher et al., 2020), which then may function as a catalyst for change in relationship quality and affective relationship description more than the reverse direction of effects.** Findings shed light on the direction and strength of effects between mothers and their **adolescent and adult** children, specifically extending findings into adolescence and adulthood in FXS.

Several previous studies identified relationships between maternal criticism and son/daughter maladaptive behaviors (Greenberg et al., 2012; Smith et al., 2016). However, consistent with Smith et al. (2016), the current study did not find an association between *change* in maternal criticism and maladaptive behaviors. The mothers in the current study produced relatively few critical remarks at both time points, and approximately 80% of the sample showed no change across 7.5 years. Thus, it is not surprising that change in maternal critical remarks was not predictive of maladaptive behaviors since minimal change occurred. Number of critical remarks during the FMSS may not be a sensitive measure of the intergenerational **affective relationship** when the **sons or daughters** are in later adolescence and/or adulthood. Additionally, it is important to note that **son/daughter** age and Time 1 critical remarks were significantly related to Time 4 critical remarks. Therefore, it may be that any variation in maternal critical remarks, albeit minimal, was more strongly associated with the **son/daughter's age** and the

mother's initial level of criticism rather than maladaptive behaviors. Future work should examine the evolution of critical remarks over time and explore other potential predictors, such as maternal mental health, parent-child relationship quality, and **son/daughter** functioning.

Significant bidirectional associations were found between maternal positive remarks and maladaptive behaviors across all behavior domains. However, importantly, change in maladaptive behaviors was a stronger predictor of Time 4 positive remarks than the reverse direction of effects. Knowing the direction of effects is critical for developing effective interventions. Supporting mothers in becoming more positive may not be as effective of a first step as intervening to change the **son/daughter**'s behavior problems first. In other words, change may need to begin at the level of the **son/daughter** when they present with clinical-level maladaptive behaviors. Sequential or simultaneous interventions that directly target maladaptive behaviors in addition to a family-based component may be more successful than family-centered interventions alone. It may be particularly difficult for mothers to increase their positivity when their **sons/daughters** have significant, often clinical-level maladaptive behaviors. Despite decreases in maladaptive behaviors across the 7.5 years, some individuals with FXS remained in the clinical range, which had downstream effects on mothers. Furthermore, one study found that approximately 31% of males and 13% of females with FXS injured their parent in the past year, at times resulting in a hospital or doctor's visit for the injury (Wheeler et al., 2016). By implementing interventions to directly address maladaptive behaviors, including pharmaceutical and behavioral treatments, mothers may ultimately be able to increase positivity toward their **son/daughter**, which may then result in further decreases in maladaptive behaviors.

Bidirectional effects were identified between asocial maladaptive behaviors and relationship quality **between the mother and her son/daughter**. The finding that asocial but not

internalized or externalized maladaptive behaviors predicted relationship quality mirrors findings in autistic adults and adolescents (Kring et al., 2010), and may be due to the greater impact of asocial behaviors (i.e., uncooperative or socially offensive behaviors) on a mother's perception of relationship quality compared to externalized or internalized behaviors, which by definition have less of a social component. Previous work suggests an association between relationship quality **between the mother and son/daughter** and maternal mental health (Fielding-Gebhardt et al., 2020), which may indicate that some mothers of **individuals** with FXS would benefit from individualized interventions to address their mental health challenges; this could potentially have positive downstream effects on relationship quality. These findings also extend previous findings from Fielding-Gebhardt et al. (2020), which identified the predictive nature of within-child change in addition to average level of maladaptive behaviors in understanding relationship quality between mothers and children with FXS during late childhood and early adolescence. As the **sons/daughters** in the current study were in late adolescence and adulthood, the findings of these two studies suggest a critical role of continued change in maladaptive behaviors in shaping relationship quality over the life course in mothers **and their sons/daughters** with FXS.

Limitations and Future Directions

While this study has many strengths, it is important to note that the original causal pathways between maternal positive remarks, relationship quality, and maladaptive behaviors began long before Time 1 of the present study. Future work should examine the direction and strength of these relationships during different developmental stages (e.g., early childhood, late adulthood) to uncover how maternal versus **son/daughter** factors may be more or less influential on the parent-child dyad at different stages of development. Moreover, it is unknown whether there is a threshold effect or an optimal level of maternal positive remarks and whether increases

in maternal positivity continue to have advantageous effects for mothers with already high levels of positive remarks. In addition, this study used the FMSS to measure maternal positivity and criticism. While this task has been used frequently in previous research and linked to **son/daughter** functioning, it is not an interaction between a mother and her **son/daughter**. Future research should examine maternal criticism and positivity during parent-child interactions. This study included mothers and their **sons/daughters** who were continuously co-residing throughout the study. Therefore, findings cannot be generalized to parent-child dyads where the adolescent or adult lives outside of the home. Furthermore, this study cannot speak to father-child relationships. The inclusion of fathers represents a critical future direction. Finally, the present study lacked racial and ethnic diversity, which is a significant limitation.

Conclusions

Adolescence and adulthood are times when many individuals with FXS show decreases in maladaptive behaviors and their mothers show increases in positivity toward their **son/daughter** with FXS, yet the stress of parenting **an individual** with FXS continues. Maternal criticism and relationship quality **between the mother and son/daughter** remain stable during this period. The relationships between maternal positive remarks, relationship quality, and maladaptive behaviors are bidirectional in nature. However, change in maladaptive behaviors may play a more substantial role in shaping maternal positivity and relationship quality than the reverse, particularly over an extended period of time. These findings lend support to interventions that directly target maladaptive behaviors and, depending on the severity of the behaviors, either simultaneously or subsequently address maternal positivity and relationship quality. While family-level interventions remain critical, improving maladaptive behaviors may require other types of direct intervention in order to induce change.

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Table 1.

Descriptive statistics and paired samples t-tests among study variables at Time 1 and Time 4

Variable	Time 1 <i>M (SD)</i>	Time 4 <i>M (SD)</i>	<i>t</i>	<i>p</i>
Maternal positive remarks	4.12 (3.13)	6.78 (4.52)	5.02	<.001
Maternal critical remarks	0.27 (0.60)	0.18 (0.42)	-1.35	.180
Relationship quality	51.23 (4.72)	51.12 (6.29)	-0.20	.843
Internalized maladaptive behaviors	112.60 (10.77)	108.82 (11.44)	-3.54	<.001
Externalized maladaptive behaviors	103.71 (9.88)	101.45 (8.35)	-2.23	.029
Asocial maladaptive behaviors	109.23 (10.66)	106.31 (10.23)	-2.39	.019

Table 2.

Standardized mean differences of maternal positive and critical remarks, relationship quality, and son/daughter maladaptive behaviors from Time 1 and Time 4 using the Time 1 standard deviation

Variable	Improved ^a		No Change ^b		Worsened ^c	
	<i>n</i>	%	<i>n</i>	%	<i>n</i>	%
Maternal positive remarks	47	60.3%	19	24.4%	12	15.4%
Maternal critical remarks	9	11.5%	63	80.8%	6	7.7%
Relationship quality	23	29.5%	32	41.0%	23	29.5%
Son/daughter internalized maladaptive behaviors	38	48.7%	28	35.9%	12	15.4%
Son/daughter externalized maladaptive behaviors	22	28.6%	44	57.1%	11	14.3%
Son/daughter asocial maladaptive behaviors	29	37.2%	37	47.4%	12	15.4%

^aImproved = Time 4 score improved compared to the Time 1 score by more than ½ SD of the Time 1 mean

^bNo Change = Time 4 score was within +/- ½ SD of the Time 1 mean

^cWorsened = Time 4 score worsened compared to the Time 1 score by more than ½ SD of the Time 1 mean

Table 3.

Results of Seemingly Unrelated Regressions (SUR) models between Time 1 and Time 4 maternal positive remarks and maladaptive behaviors

	Unstandardized coefficient (β)	Standard error	<i>p</i> -value	<i>R</i> ²
<i>SUR Model 1: Maternal Positive Remarks & Internalized Maladaptive Behaviors</i>				
Multiple regression predicting Time 4 internalized maladaptive behaviors				
Intercept	38.94	11.12	.001**	.478
Time 1 internalized maladaptive behaviors	0.70	0.09	<.001***	
Time 1 maternal positive remarks	-1.17	0.32	.001**	
Time 4 – Time 1 maternal positive remarks	-1.53	0.21	<.001***	
Son/daughter age	-0.01	0.14	.960	
Multiple regression predicting Time 4 maternal positive remarks				
Intercept	14.17	5.78	.015*	.177
Time 1 maternal positive remarks	0.34	0.15	.025*	
T1 internalized maladaptive behaviors	-0.09	0.05	.052	
Time 4 – Time 1 internalized maladaptive behaviors	-0.35	0.05	<.001***	
Son/daughter age	0.00	0.07	.993	
<i>SUR Model 2: Maternal Positive Remarks & Externalized Maladaptive Behaviors</i>				
Multiple regression predicting Time 4 externalized maladaptive behaviors				
Intercept	67.39	8.71	<.001***	.339
Time 1 externalized maladaptive behaviors	0.41	0.08	<.001***	
Time 1 maternal positive remarks	-0.82	0.27	.003**	
Time 4 – Time 1 maternal positive remarks	-0.64	0.19	.001**	
Son/daughter age	-0.20	0.12	.094	
Multiple regression predicting Time 4 maternal positive remarks				
Intercept	24.08	6.96	.001**	.096
Time 1 maternal positive remarks	0.31	0.16	.060	
T1 externalized maladaptive behaviors	-0.18	0.06	.005**	
Time 4 – Time 1 externalized maladaptive behaviors	-0.23	0.07	.001**	
Son/daughter age	-0.03	0.08	.656	
<i>SUR Model 3: Maternal Positive Remarks & Asocial Maladaptive Behaviors</i>				

Multiple regression predicting Time 4 asocial maladaptive behaviors				
Intercept	60.97	11.33	<.001***	.230
Time 1 asocial maladaptive behaviors	0.46	0.09	<.001***	
Time 1 maternal positive remarks	-0.58	0.36	.110	
Time 4 – Time 1 maternal positive remarks	-0.95	0.25	<.001***	
Son/daughter age	0.01	0.15	.943	
Multiple regression predicting Time 4 maternal positive remarks				
Intercept	16.74	6.19	.008**	.093
Time 1 maternal positive remarks	0.42	0.16	.009**	
T1 asocial maladaptive behaviors	-0.11	0.05	.035*	
Time 4 – Time 1 asocial maladaptive behaviors	-0.20	0.05	<.001***	
Son/daughter age	0.01	0.15	.943	

Note. * $p < .050$, ** $p < .010$, *** $p < .001$. Bolded indicates the change variable associated with each multiple regression model.

Table 4.

Results of Seemingly Unrelated Regressions (SUR) models between Time 1 and Time 4 maternal critical remarks and maladaptive behaviors

	Unstandardized coefficient (β)	Standard error	<i>p</i> -value	<i>R</i> ²
<i>SUR Model 4: Maternal Critical Remarks & Internalized Maladaptive Behaviors</i>				
Multiple regression predicting Time 4 internalized maladaptive behaviors				
Intercept	33.84	11.23	.003**	.424
Time 1 internalized maladaptive behaviors	0.66	0.09	<.001***	
Time 1 maternal critical remarks	-0.60	2.54	.816	
Time 4 – Time 1 maternal critical remarks	-3.82	2.63	.148	
Son/daughter age	0.02	0.16	.903	
Multiple regression predicting Time 4 maternal critical remarks				
Intercept	0.23	0.51	.657	.214
Time 1 maternal critical remarks	0.28	0.07	<.001***	
T1 internalized maladaptive behaviors	0.00	0.00	.338	
Time 4 – Time 1 internalized maladaptive behaviors	-0.01	0.00	.148	
Son/daughter age	0.02	0.01	.014*	
<i>SUR Model 5: Maternal Critical Remarks & Externalized Maladaptive Behaviors</i>				
Multiple regression predicting Time 4 externalized maladaptive behaviors				
Intercept	57.07	8.90	<.001***	.309
Time 1 externalized maladaptive behaviors	0.46	0.08	<.001***	
Time 1 maternal critical remarks	-0.97	2.12	.648	
Time 4 – Time 1 maternal critical remarks	-0.33	2.14	.876	
Son/daughter age	-0.17	0.12	.167	
Multiple regression predicting Time 4 maternal critical remarks				
Intercept	-0.73	0.58	.208	.227
Time 1 maternal critical remarks	0.24	0.07	.001**	
T1 externalized maladaptive behaviors	0.01	0.01	.353	
Time 4 – Time 1 externalized maladaptive behaviors	0.00	0.01	.876	
Son/daughter age	0.02	0.01	.011*	
<i>SUR Model 6: Maternal Critical Remarks & Asocial Maladaptive Behaviors</i>				

Multiple regression predicting Time 4 asocial maladaptive behaviors				
Intercept	55.55	11.01	<.001***	.221
Time 1 asocial maladaptive behaviors	0.46	0.10	<.001***	
Time 1 maternal critical remarks	-2.64	2.65	.320	
Time 4 – Time 1 maternal critical remarks	-4.68	2.73	.088	
Son/daughter age	0.06	0.16	.718	
Multiple regression predicting Time 4 maternal critical remarks				
Intercept	-0.02	0.52	.969	.215
Time 1 maternal critical remarks	0.27	0.07	<.001***	
T1 asocial maladaptive behaviors	0.00	0.00	.668	
Time 4 – Time 1 asocial maladaptive behaviors	-0.01	0.00	.088	
Son/daughter age	0.02	0.01	.010*	

Note. * $p < .050$, ** $p < .010$, *** $p < .001$. Bolded indicates the change variable associated with each multiple regression model.

Table 5.

Results of Seemingly Unrelated Regressions (SUR) models between Time 1 and Time 4 relationship quality and maladaptive behaviors

	Unstandardized coefficient (β)	Standard error	<i>p</i> -value	<i>R</i> ²
<i>SUR Model 7: Relationship Quality & Internalized Maladaptive Behaviors</i>				
Multiple regression predicting Time 4 internalized maladaptive behaviors				
Intercept	47.92	16.50	.004**	.421
Time 1 internalized maladaptive behaviors	0.69	0.09	<.001***	
Time 1 relationship quality	-0.32	0.21	.141	
Time 4 – Time 1 relationship quality	-0.35	0.20	.080	
Son/daughter age	-0.01	0.15	.969	
Multiple regression predicting Time 4 relationship quality				
Intercept	6.65	9.82	.499	.365
Time 1 relationship quality	0.76	0.12	<.001***	
T1 internalized maladaptive behaviors	0.03	0.06	.553	
Time 4 – Time 1 internalized maladaptive behaviors	-0.11	0.06	.080	
Son/daughter age	0.06	0.09	.484	
<i>SUR Model 8: Relationship Quality & Externalized Maladaptive Behaviors</i>				
Multiple regression predicting Time 4 externalized maladaptive behaviors				
Intercept	66.18	14.02	<.001***	.310
Time 1 externalized maladaptive behaviors	0.45	0.08	<.001***	
Time 1 relationship quality	-0.15	0.18	.387	
Time 4 – Time 1 relationship quality	-0.19	0.16	.219	
Son/daughter age	-0.16	0.12	.174	
Multiple regression predicting Time 4 relationship quality				
Intercept	9.89	11.39	.387	.359
Time 1 relationship quality	0.79	0.13	<.001***	
T1 externalized maladaptive behaviors	0.00	0.08	1.00	
Time 4 – Time 1 externalized maladaptive behaviors	-0.10	0.08	.219	
Son/daughter age	0.03	0.09	.757	
<i>SUR Model 9: Relationship Quality & Asocial Maladaptive Behaviors</i>				
Multiple regression predicting Time 4 asocial maladaptive behaviors				
Intercept	76.19	17.62	<.001***	.230
Time 1 asocial maladaptive behaviors	0.42	0.10	<.001***	

Time 1 relationship quality	-0.33	0.22	.148	
Time 4 – Time 1 relationship quality	-0.77	0.19	<.001***	
Son/daughter age	0.03	0.15	.869	
Multiple regression predicting Time 4 relationship quality				
Intercept	28.13	10.43	.008**	.356
Time 1 relationship quality	0.73	0.12	<.001***	
T1 asocial maladaptive behaviors	-0.14	0.06	.024*	
Time 4 – Time 1 asocial maladaptive behaviors	-0.24	0.06	<.001***	
Son/daughter age	0.04	0.08	.598	

Note. * $p < .050$, ** $p < .010$, *** $p < .001$. Bolded indicates the change variable associated with each multiple regression model.