

Title: Predictors of Maternal Well-Being and Adolescent and Young Adult Behavior in Fragile X Families

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Introduction: Fragile X syndrome (FXS) is a genetic disorder caused by a single gene mutation on the X chromosome and is the leading inherited cause of intellectual disability (Tassone et al., 2012). Phenotypically, individuals with FXS often exhibit various challenging behaviors such as anxiety, hyperactivity, repetitive/restricted interests, and attention problems (Smith et al., 2012). Indeed, the presence of these types of challenging behaviors can have a negative impact on family outcomes, in particular maternal well-being (Hauser, Kover & Abbeduto, 2014). In addition, as carriers of either the *FMR1* full or premutation, mothers of children with FXS are predisposed to present with mental health concerns such as anxiety and depression (Gossett et al., 2016) in addition to increased levels of parenting stress. However, there is still relatively little research addressing the reciprocal nature of the association between youth challenging behaviors and maternal well-being and the potential impact that other factors such as family economic status might have on maternal well-being. Understanding these types of reciprocal relationships is vital for supporting optimal outcomes in both youth with FXS and their caregivers. In the present study, we examined (1) how maternal and youth characteristics contribute to maternal well-being one year later and (2) potential carry over effects of aspects of maternal well-being on youth behavior one year later.

Method: Data collected in a longitudinal study on 35 males with FXS between the ages of 15 and 22 years, and their biological mothers, were used in the present project. The severity of challenging behaviors in the youth with FXS was measured using the ADAMS Behavior Scale, a 28-item parent report screener for the presence/severity of challenging behaviors that yields five subscale scores (i.e., Manic/Hyperactive Behavior, Depressed Mood, Social Avoidance, General Anxiety, and Obsessive/Compulsive Behavior). To evaluate overall maternal well-being, aspects of parenting stress, anxiety, and satisfaction with life were assessed using the Parenting Stress Inventory (PSI-4), the Beck Anxiety Index (BAI), and the Satisfaction With Life Scale (SWLS) respectively. The PSI-4 is a 36 item self-assessment that measures parenting distress, parent-child dysfunctional interaction, difficult child, and total parenting stress. The BAI is a 21 item self-report inventory that measures anxiety severity, and the SWLS is a multi-item scale to measure overall life satisfaction. Further, we considered maternal age, educational attainment, full-scale IQ as measured by the KBIT-2, and parent report of family income, as additional maternal characteristics which might predict maternal well-being.

Results: A series of multiple linear regressions were computed to look at the relationship between both maternal and youth characteristics on subsequent maternal well-being. In the first series of models we looked at the predictive nature of maternal education, cognitive ability, age, and family income at time one on maternal well-being measured one year later (e.g., parenting stress, anxiety, and overall satisfaction with life). Overall regression models were significant, with family income being the only unique predictor of parental distress, total parenting stress, and satisfaction of life. The next series of regression models considered ratings of youth challenging behavior at time one (i.e., manic/hyperactivity, depression, social avoidance, general anxiety, and obsessive/compulsive behaviors), on aspects of maternal well-being measured one year later. Results indicated that youth challenging behavior predicted parenting stress and satisfaction of life, with youth obsessive/compulsive behaviors emerging as a unique predictor of parental distress, total parenting stress, and satisfaction of life. Finally, we completed another series of multiple linear regressions to address whether factors of maternal well-being, after accounting for youth challenging behavior measured concurrently, predicted ratings of youth challenging behavior measured one year later. In addition to the predictive associations observed between the ratings of youth challenging behaviors across time, parental distress, difficult child, and total parenting stress were also significant predictors of youth depression.

Discussion: The purpose of this study was to examine potential predictors of maternal well-being and subsequent effects on adolescent/young adult challenging behavior. We found that maternal characteristics, in particular family income and youth obsessive/compulsive behaviors, were indeed predictors of maternal self-report of parenting stress and satisfaction of life. Further, we found preliminary support for a reciprocal relationship between maternal well-being and ratings of challenging

behaviors in adolescent/young adults with FXS. Additionally, our findings highlight the impact that family income has on multiple aspects of maternal well-being, showing that further work is needed to expand access to services to families in lower income and rural areas. One such way of accomplishing this is through the use of distance technology to limit costs, including time and financial, associated with travel. Importantly, these findings can help suggest ways to better support this population of families in terms of future teaching and therapy techniques.

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