

Title: Parental Well-Being and Family Relationships in Families Affected by Fragile X Syndrome

Authors: Sarah Nelson Potter & Leonard Abbeduto – UC Davis MIND Institute

Introduction: Given the phenotypic characteristics of children with fragile X syndrome (FXS), mothers and fathers of children with FXS are likely to experience heightened levels of parenting stress (PS) (Hartley, Seltzer, Head, & Abbeduto, 2012; McCarthy, Cuskelly, van Kraayenoord, & Cohen, 2006), which may impede their ability to engage with their children in the sustained and responsive interactions that facilitate development. Elevated PS may also negatively affect the marital relationship, which may further exacerbate problems in parent-child interactions (Johnston et al., 2003). Furthermore, mental health challenges (MHC), which are common among women with the *FMR1* pre- or full mutation (Hagerman et al., 2018), may be exacerbated by the stress of raising a child with significant challenges (Hagerman & Hagerman, 2004). MHC are also associated with decreased marital cohesion and couple satisfaction (Baker, Seltzer, & Greenberg, 2012). Overall, poor parental and marital functioning contribute to negative outcomes in typically developing children (Masarik & Conger, 2017). The present study was designed to examine parental well-being and marital relationships in families affected by FXS as well as the influence of child characteristics on parental and couple well-being.

Method: Data collection is ongoing and approximately 30 couples who have a 3 to 7-year-old male child with FXS will participate by March of 2021. Seventeen families have been recruited as of October 2020 with data from nine families included in the current analyses. Data are being collected entirely in family homes via distance using video conferencing and online surveys. Each parent is independently completing multiple questionnaires including the Symptom Checklist-90-Revised (SCL-90-R) as a measure of MHC, the Parenting Stress Index-4th Edition-Short Form (PSI-4-SF) as a measure of PS, the Parenting Sense of Competence Scale (PSOC) as a measure of parenting satisfaction and efficacy, the Couples Satisfaction Index (CSI) as a measure of couples satisfaction/cohesion, the Dyadic Coping Inventory (DCI) as a measure of parents' coping skills, the Aberrant Behavior Checklist-2nd Edition (ABC-2) as a measure of child challenging behaviors, and the Social Responsiveness Scale-2nd Edition (SRS-2) as a measure of child autism spectrum disorder (ASD) symptoms. One parent in each couple is also completing the Vineland-3 interview as a proxy for child developmental level.

Results: Five of nine mothers and five of nine fathers reported clinically significant levels of MHC on the SCL-90-R. Furthermore, seven mothers and four fathers reported clinically significant levels of PS in the Difficult Child domain on the PSI-4-SF. Interspousal correlations between parents' ratings of MHC and PS indicated significant correspondence between their scores on the SCL-90-R in the dimensions of Anxiety and Paranoid Ideation and on the PSI-4-SF in the Parent-Child Dysfunctional Interaction and Total Stress domains. Despite both mothers and fathers endorsing relatively high levels of MHC and PS, both groups reported that they were satisfied in their marital relationships and indicated normative levels of parenting satisfaction/efficacy and dyadic coping. For mothers, there were significant correlations between scores on the SCL-90-R and the PSOC, between the PSI-4-SF and both the PSOC and CSI, and between the CSI and DCI. For fathers, there were significant correlations between scores on the SCL-90-R and both the PSI-4-SF and PSOC, between the PSI-4-SF and PSOC, and between the PSOC and both the CSI and DCI. Furthermore, on average, mothers reported higher levels of child challenging behaviors on the ABC-2 compared to fathers and maternal and paternal ratings were not significantly correlated. Seven mothers and six fathers reported clinically significant levels of child ASD symptoms on the SRS-2, with three mothers and four fathers endorsing scores that fell in the severe range. Interspousal correlations were significant for the SRS-2. Additionally, for mothers, the ABC-2 was significantly correlated only with the CSI, whereas the SRS-2 was significantly correlated with both the PSI-4-SF and the PSOC. For fathers, the ABC-2 was significantly correlated with the SCL-90-R, the PSI-4-SF, and the PSOC, and the SRS-2 was significantly correlated with the SCL-90-R, the PSOC, and the ABC-2. All of the foregoing significant correlations were in the hypothesized direction.

Discussion: Unfortunately, many mothers and fathers of children with FXS reported clinically significant levels of MHC and PS, suggesting that they need more support. However, despite these challenges, most mothers and fathers reported moderate to

high levels of parenting satisfaction and efficacy, couples satisfaction, and dyadic coping, which may be protective against the daily stressors that they are encountering. Overall, mothers compared to fathers tended to report higher levels of child challenging behaviors and symptoms of ASD, which may be related to the fact that the majority of mothers in the present study identified themselves as the child's primary caretaker and were thus, more familiar with the child's challenges. However, interestingly, fathers' ratings of child challenging behaviors and symptoms of ASD were more strongly associated with their individual and couple well-being scores compared to mothers' ratings, suggesting that fathers may be affected by child characteristics to a greater extent than mothers, which is consistent with past research (Hartley, Barker, Seltzer, Greenberg, & Floyd, 2011; McCarthy et al., 2006). Including parents in behavioral therapies and health care services is critical to a child's success, especially for young children. In order to maximize treatment gains for children with FXS and to improve well-being for the entire family system, we need to consider the well-being of mothers and fathers. This study provides us with a greater understanding of the needs of, and challenges faced by, families affected by FXS, which will allow us to better tailor the services and interventions that we provide.

References:

- Baker, J. K., Seltzer, M. M., & Greenberg, J. S. (2012). Behaviour problems, maternal internalising symptoms, and family relations in families of adolescents and adults with fragile X syndrome. *Journal of Intellectual Disability Research*, 56(10), 984-995.
- Hagerman, P. J., & Hagerman, R. J. (2004). The fragile-X premutation: A maturing perspective. *The American Journal of Human Genetics*, 74(5), 805-816.
- Hagerman, R. J., Protic, D. D., Rajaratnam, A., Salcedo-Arellano, M. J., Aydin, E. Y., & Schneider, A. (2018). Fragile X-Associated Neuropsychiatric Disorders (FXAND). *Frontiers in Psychiatry*, 9, 1-9.
- Hartley, S. L., Barker, E. T., Seltzer, M. M., Greenberg, J. S., & Floyd, F. J. (2011). Marital satisfaction and parenting experiences of mothers and fathers of adolescents and adults with autism. *American Journal of Intellectual and Developmental Disabilities*, 116(1), 81-95.
- Hartley, S. L., Seltzer, M. M., Head, L., & Abbeduto, L. (2012). Psychological well-being in fathers of adolescents and young adults with Down Syndrome, Fragile X syndrome, and autism. *Family Relations*, 61(2), 327-342.
- Johnston, C., Hessel, D., Blasey, C., Eliez, S., Erba, H., Dyer-Friedman, J., ... & Reiss, A. L. (2003). Factors associated with parenting stress in mothers of children with fragile X syndrome. *Journal of Developmental & Behavioral Pediatrics*, 24(4), 267-275.
- Masarik, A. S., & Conger, R. D. (2017). Stress and child development: A review of the family stress model. *Current Opinion in Psychology*, 13, 85-90.
- McCarthy, A., Cuskelly, M., van Kraayenoord, C. E., & Cohen, J. (2006). Predictors of stress in mothers and fathers of children with fragile X syndrome. *Research in Developmental Disabilities*, 27(6), 688-704.