Title: Trajectories of Change in the Behavioral and Health Phenotype of Adolescents and Adults with Fragile X Syndrome and Intellectual Disability: Longitudinal Trends over a Decade

Authors: Lauren V. Usher¹, Leann S. DaWalt¹, Jan S. Greenberg¹, Marsha R. Mailick¹
²University of Wisconsin-Madison

Introduction: Individuals with fragile X syndrome (FXS) display behavior problems, functional limitations, and health problems, across the lifespan (Smith et al., 2016; Raspa et al., 2010). However, much of what we know about the phenotype comes from cross-sectional studies investigating children and adolescents. In this study, longitudinal data was utilized to examine trajectories of behavior (daily living skills and behavior problems) and health (body mass index [BMI] and health rating) in adolescents and adults with FXS, accounting for age, sex, and autism spectrum disorder (ASD) symptoms.

Method: Participants (n=134) were drawn from a longitudinal study of families of adolescents and adults with FXS (Mailick et al., 2014). At study entry, individuals with FXS met three criteria: 1) were 12 years of age or older, 2) had received a diagnosis of FXS, and 3) lived with parents or had at least weekly contact. If there was more than one co-residing child, mothers reported on the child she viewed as most severely affected. Individuals with FXS were mostly males (85.1%), in their 20s on average (M = 20.19, SD = 6.92, range 12–48 years), and most lived with mothers (90.3%). About one-quarter (24.2%) of the sample had a co-occurring diagnosis of ASD. Mothers reported on sons’ or daughters’ daily living skills, behavior problems, height and weight, and health, as well as lifetime ASD symptoms. The present sample was restricted to those with intellectual disability.

Results: Hierarchical linear modeling was used to describe change in the behavioral and health phenotype of adolescents and adults over ten years and to examine effects of between-person differences on trajectories. We evaluated separate growth curve models for four outcomes: daily living skills, behavior problems, BMI, and health. Age, sex, and lifetime ASD symptoms at Time 1 were entered as predictors of the initial time point (intercept) and the linear and quadratic slopes. Results indicated that adaptive skills, behavior problems, and health rating improved over time. However, BMI increased over time, indicating a greater proportion of individuals meeting the cutoffs for overweight or obesity across the course of the study. At baseline, adolescents and adults with fewer ASD symptoms were more independent in daily living skills, in better health, and had fewer behavior problems. People who were older at study entry were more independent in daily living skills but had higher BMI than younger people. Also, older people changed at a slower rate with respect to daily living skills, behavior problems, and BMI.

Discussion: This is the first study to examine longitudinal trajectories of behavioral and health phenotypes for adolescents and adults with FXS over the course of a decade. Trajectories for daily living skills and behavior problems indicated improved over time, with greater change for those at younger ages. Regarding health, change was greater for females than for males. Individuals with elevated levels of ASD symptoms were at risk for more behavior problems and poorer health at the initial time point than were those with lower levels of ASD symptoms, suggesting possible targets for intervention.

References/Citations:


Acknowledgements:

- This work was supported by the National Institute of Child Health and Human Development (T32 HD07489, Hartley, PI, and R01 HD082110, Mailick, PI) and the Waisman Center IDDRC (U54 HD090256, Chang, PI).