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A comparison of functional academic and daily living skills in males with fragile X syndrome with and without autism

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ABSTRACT

Background: Adaptive behaviors, such as functional academic and daily living skills, are critical for independence in adults with intellectual and developmental disabilities. However, little is known about these skills in fragile X syndrome (FXS), the most common form of inherited intellectual disability.

Aims: The purposes of this study were to describe the functional academic and daily living skills of males diagnosed with FXS across different age groups and compare skill attainment by autism status and other common co-occurring conditions.

Methods and procedures: We used survey methods to assess parent-reported functional academic and daily living skills in 534 males with FXS. Functional academic skills included time and schedules, money, math, reading, and writing skills. Daily living skills included hygiene, cooking, laundry and housekeeping, transportation, and safety skills.

Outcomes and results: Analyses examined functional academic and daily living skills in a cross-sectional sample of males between ages 5 and 67. Differences in skill attainment were found by child age, co-morbid autism status, total number of co-occurring conditions, and respondent education. Functional academic and daily living skills were predictive of community employment and independent living.

Conclusions and implications: These data provide important information on the mastery of both foundational and more complex adaptive skills in males with FXS. Both functional academic and daily living skills were predictive of measures of independence above and beyond other child and family characteristics. These findings point to the need to focus interventions to support the attainment of independence in males with FXS.

What this paper adds

This study provides additional evidence of the range of functional academic and daily living skills in males with FXS. In keeping with prior research, the results indicate that these adaptive behaviors develop over time and are related to other co-morbid conditions, such as autism. Males attained foundational skills, such as counting to 10 and writing own name, earlier than more complex skills, such as knowing the value of coins and small bills or writing words from memory. Many adult males showed independent daily living skills related to hygiene, preparing simple meals, and basic housekeeping skills. Adaptive skills were the strongest predictor of employment status and living arrangement in adult males with FXS.

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1. Introduction

Fragile X syndrome (FXS) is a single-gene, X-linked disorder and the most common form of inherited intellectual disability (Berry-Kravis, Grossman, Crnic, & Greenough, 2002; Huddleston, Visootsak, & Sherman, 2014; Raspa, Wheeler, & Riley, 2017). In the general population, FXS occurs in approximately 1 in 4000 males (Crawford, Acuna, & Sherman, 2001; Hagerman et al., 2009). Individuals with FXS often have difficulties with cognitive functions such as attention (Cornish, Cole, Longhi, Karmiloff-Smith, & Scerif, 2013), executive functions (Hooper et al., 2008), memory (Ornstein et al., 2008), language (Abbeduto, Brady, & Kover, 2007; Martin, Losh, Estigarribia, Sideris, & Roberts, 2013), and learning (Buckley & Johnson-Glenberg, 2008). Other core features of FXS include anxiety (Cordeiro, Ballinger, Hagerman, & Hessl, 2011; Wheeler et al., 2014) and hyperarousal (Hall, Lightbody, Huffman, Lazzeroni, & Reiss, 2009). Behavior challenges, such as aggression (Wheeler, Raspa, Bishop, & Bailey, 2016) or self-injury (Symons, Byiers, Raspa, & Bishop, 2010), are seen in a subset of individuals with FXS as well. Up to one-third or more of males with FXS are co-diagnosed with autism spectrum disorder (ASD) (Bailey, Mesibov, Hatton, Clark, & Roberts, 1998; Hatton et al., 2006; Hernandez et al., 2009; Kaufmann et al., 2004).

The co-occurrence of autism defines a sub-phenotype of FXS. Individuals with comorbid FXS and ASD exhibit poorer developmental outcomes, including weaker communication and social skills, increased number of behavior problems, and greater cognitive impairment than males with FXS only or individuals with idiopathic autism (Bailey, Hatton, Mesibov, Ament, & Skinner, 2000; Kaufmann et al., 2004; Reisinger & Roberts, 2017). Individuals with FXS and co-morbid ASD also have deficits in social behaviors and skills (Lewis et al., 2006; Smith, Barker, Seltzer, Abbeduto, & Greenberg, 2012) or increases in social withdrawal (Budimirovic et al., 2006; Kau et al., 2004) when compared to those with FXS only. Indeed, social impairments may be the most significant predictor that differentiates individuals with co-morbid FXS and ASD from FXS only (Brock & Hatton, 2010).

Although understanding how FXS affects cognitive and behavioral functioning is important, having a better understanding of an individual's adaptive skills is critical as well. Adaptive behaviors are used in everyday life and include functional academic skills, social and communication skills, and daily living skills. Higher levels of adaptive behaviors have been shown to be associated more independence in adulthood whereas lower levels of adaptive skills often require more use of community supports (Woolf, Woolf, & Oakland, 2010). In accordance with the *International Classification of Functioning, Disability, and Heath* (ICF) (WHO, 2001), assessment of adaptive skills can be used to determine level of impairment and can be used to plan for interventions in family, school, or social contexts. Having a deeper knowledge of adaptive skills in FXS also provides baseline information which can be used to evaluate the results of educational, therapeutic, or social interventions and treatments.

Limited research has been conducted on adaptive behaviors in FXS. An early study showed both difficulties and strengths in adaptive skills as measured by the Vineland Adaptive Behavior Scales (Dykens, Hodapp, Ort, & Leckman, 1993). Individuals with FXS had relative strengths in personal (e.g., hygiene) and domestic (e.g., housekeeping) skills, but had more difficulty with community skills (e.g., managing money). More recently, Bailey, Raspa, Holiday, Bishop, and Olmsted (2009) used data from a national survey of parents of 1105 individuals with FXS and showed that the majority of adults master many basic self-care skills such as toileting, dressing, eating, and bathing. More advanced functional academic skills, such as using complex sentences and reading, were less likely to be achieved. Is a cross-sectional analysis, the study also found a steady increase in the percent of individuals who had mastered these adaptive skills in the early childhood years, with tapering off in adolescence and adulthood. However, two longitudinal studies of individuals with FXS showed declines in adaptive behaviors over time, including daily living skills, communication, and socialization (Fisch, Simensen, & Schroer, 2002; Klaiman et al., 2014). Predictors of adaptive behavior in individuals with FXS include gender, age, and IQ (Bailey et al., 2009; Glaser et al., 2003; Klaiman et al., 2014). Individuals with FXS and comorbid ASD exhibit greater delays in adaptive functioning when compared with those with FXS only (Hatton et al., 2003; Kau et al., 2004; Kaufmann et al., 2004).

Given the importance of adaptive behaviors and the limited data on functional academic and daily living skills, the purposes of this study are to (a) describe the functional academic and daily living skills of males diagnosed with FXS across different age groups, (b) compare the functional academic skills and daily living skills of males with and without ASD as well as other covariates, (c) examine whether functional academic and daily living skills are related to measures of independence, including living arrangement and employment type.

2. Method

2.1. Study design

The data used for this study were part of a large, U.S. national fragile X family survey. The survey included several modules which focused on a range of topics related to the FXS phenotype, such as social skills, autism symptoms, functional academic and daily living skills, medication and treatment, and family impact. Families were recruited from a survey research registry, *Our Fragile X World*. The majority of participants (94%) completed the survey online and a few by telephone (6%). Data collection for the survey began in spring, 2012 and lasted approximately 6 months. The study was reviewed and approved by an Institutional Review Board prior to implementation. At the start of the survey, family participants were given a consent form to read and acknowledge before responding to any questions.