



## Examining the operant function of challenging behavior in young males with fragile X syndrome: A summary of 12 cases



Wendy Machalicek<sup>a,\*</sup>, Andrea McDuffie<sup>b</sup>, Ashley Oakes<sup>b</sup>, Monica Ma<sup>b</sup>,  
Angela John Thurman<sup>b</sup>, Mandy J. Rispoli<sup>c</sup>, Leonard Abbeduto<sup>b</sup>

<sup>a</sup> University of Oregon, United States

<sup>b</sup> University of California-Davis MIND Institute, United States

<sup>c</sup> Texas A&M University, United States

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### ABSTRACT

This study used experimental functional analyses to examine the operant function of challenging behaviors exhibited by 12 males (ages 27–51 months) with fragile X syndrome (FXS). Eight children met criteria for negatively reinforced challenging behavior in the form of escape from demands and/or escape from social interactions. Nine children met criteria for positively reinforced challenging behavior in the form of obtaining access to highly preferred items. Attention was identified as a maintaining consequence for three children. The functional analysis was inconclusive for one child. Results suggest that, for young males with FXS, challenging behaviors may more likely be tangibly and escape maintained than attention maintained. Our findings affirm past research suggesting a unique behavioral phenotype for this population.

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## 1. Introduction

Fragile X syndrome (FXS) is the leading known cause of inherited intellectual disability (Crawford, Acuna, & Sherman, 2001). The etiology of FXS is a mutation in the *FMR1* gene, which lies on the long arm of the X chromosome at Xq27.3 (Verkerk et al., 1991). Expression of the syndrome is dependent on the number of CGG nucleotide repeats with more than 200 repeats resulting in full mutation and a high probability of hyper-methylation of the promoter region of the *FMR1* gene. A full mutation and hyper-methylation of the *FMR1* gene stops production of the fragile X mental retardation protein (FMRP), which is involved in synaptic plasticity and experience-dependent learning (Bassall & Warren, 2008; Taylor et al., 1994).

In addition to cognitive deficits, individuals with FXS often display a variety of challenging behaviors including: hyperactivity and attentional difficulties (Cornish, Scerif, & Karmiloff-Smith, 2007; Scerif, Longhi, Cole, Karmiloff-Smith, & Cornish, 2012; Turk, 1998; Baumgardner, Reiss, Freund, & Abrams, 1995); anxiety and withdrawal (Bregman, Leckman, & Ort, 1988; Cordeiro, Ballinger, Hagerman, & Hessler, 2011; Kau, Reider, Payne, Meyer, & Freund, 2000); repetitive behaviors (Hall, Lightbody, Hirt, Rezvani, & Reiss, 2010); self-injurious behaviors (Hagerman & Hagerman, 2002; Hall, Lightbody, & Reiss, 2008; Symons, Clark, Hatton, Skinner, & Bailey, 2003); and aggression (Leigh, Hagerman, & Hessler, 2012). Such

\* Corresponding author at: Department of Special Education and Clinical Sciences, University of Oregon, 901 East 18th Avenue, Eugene, OR 97403, United States. Tel.: +1 541 346 4404; mobile: +1 608 320 6028.

E-mail address: [wmachali@uoregon.edu](mailto:wmachali@uoregon.edu) (W. Machalicek).

behaviors are especially prevalent in males with FXS (Hagerman & Hagerman, 2002; Hall et al., 2008; Symons et al., 2003) with self-injury reported to occur in 58% of boys and 17% of girls with FXS and compulsive behavior occurring in 72% of boys and 55% of girls with FXS (Hall et al., 2008). Additionally, as many as 60% of males with FXS display behaviors that are frequent and severe enough to warrant a diagnosis of autism spectrum disorder (ASD) (Bailey et al., 1988; Demark, Feldman, & Holden, 2003; Hagerman et al., 1986; Hall et al., 2010; Harris et al., 2008; Hatton et al., 2006; Lewis et al., 2006; Sabaratnam, Murthy, Wijeratne, Buckingham, & Payne, 2003). Individuals with comorbid diagnoses of FXS and ASD typically experience higher rates of challenging behaviors than individuals diagnosed with idiopathic ASD or FXS alone (Smith, Barker, Seltzer, Abbeduto, & Greenberg, 2012).

Research suggests that individuals with FXS may present a unique behavioral pattern that differs from individuals with other intellectual and developmental disabilities. Specifically, individuals with FXS may be more likely to engage in challenging behavior to obtain preferred tangibles and to escape demands (Hall, DeBernadis, & Reiss, 2006; Symons et al., 2003), but they may be less likely to engage in challenging behavior to obtain adult attention (Langthorne & McGill, 2012; Symons et al., 2003; Woodcock, Oliver, & Humphreys, 2009). Symons et al. (2003) found that 87% of parents of boys with FXS reported that their child engaged in challenging behaviors following changes in routines and 65% of parents reported task demands to precede challenging behavior. Only 3% of parents reported that their child engaged in challenging behaviors to access adult attention. To date, however, only a single study has examined these findings using functional analysis methodology (Langthorne et al., 2011).

Functional analysis is the only type of functional behavior assessment that makes it possible to prove a causal relationship between social consequences and challenging behavior (Hanley, Iwata, & McCord, 2003; Iwata & Dozier, 2008). In functional analysis of the challenging behavior demonstrated by 8 children ages 8–15 years with FXS, Langthorne et al. (2011) found that no child met criteria for attention-maintained challenging behavior, 5 children met the criteria for escape-maintained challenging behavior, and 4 children met the criteria for tangible-maintained challenging behavior, consistent with the parent report data of Symons et al. (2003).

Assessing the operant function of challenging behavior in young children with FXS is important as previous research indicates that challenging behaviors often appear very early in males with full mutation FXS (Bailey, Raspa, Olmstead, & Holiday, 2008; Symons, Byiers, Raspa, Bishop, & Bailey, 2010). Additionally, a growing body of research demonstrates that even the challenging behaviors associated with genetic syndromes such as FXS are sensitive to environmental antecedents and consequences (Anderson, Dancis, & Alpert, 1978; Hall et al., 2006; Hall, Oliver, & Murphy, 2001; Lesniak-Karpiak, Mazzocco, & Ross, 2003; Oliver, Murphy, Crayton, & Corbett, 1993; Sloneem, Arron, Hall, & Oliver, 2009; Symons et al., 2010; Taylor & Oliver, 2008). Using functional analysis methodology to determine the operant function of challenging behaviors in younger children with FXS will be important because it may lead to the development of effective early behavioral intervention, thus preventing the worsening of behaviors, as the child grows older.

In the current study, we implemented functional analysis procedures similar to that described by Iwata, Dorsey, Slifer, Bauman, and Richman (1982–1994) with a group of 12 young children, ages 27–51 months, with full mutation FXS and cognitive and language delays. The biological mothers of each child implemented functional analysis procedures with their child with the support of a clinician. The results of the functional analysis were compared to indirect functional behavior assessment results obtained through maternal completion of the Questions About Behavioral Function (QABF; Matson & Vollmer, 1995). The goals were to examine the social consequences maintaining challenging behaviors and to evaluate the degree of correspondence between parent report (assessed by the QABF) and the experimental functional analysis.

## 2. Methods

### 2.1. Participant characteristics and target behaviors

Twelve children and their biological mothers provided informed consent prior to participating in the current study, which was approved by the authors' university ethics review boards and carried out in line with the 1964 Declaration of Helsinki and subsequent revision (Williams, 2008). Families throughout the United States and Canada were recruited through a website posting by the National Fragile X Foundation for a larger study evaluating the effects of a parent-implemented naturalistic communication and behavioral intervention. Inclusion criteria for the children were: (a) male gender; (b) diagnosis of the FXS full mutation documented through molecular genetic testing; (c) use of fewer than 5 different spoken words according to parent report, but displaying at least one intentional communicative act during administration of the Autism Diagnostic Observation Schedule-Second Edition (ADOS-2; Lord et al., 2012); and (d) the absence of serious sensory or motor impairments according to parent report.

Each child's mother reported a variety of challenging behaviors during an intake interview conducted by the second author using an adapted form of the Routines Based Interview (RBI; McWilliam, 2003). The adapted RBI included (a) a section for the interviewer to document challenging behaviors during discussed routines and (b) follow-up questions from O'Neill et al. (1997) functional behavior assessment interview when the mother reported challenging behavior. In addition, each child was observed during baseline assessment sessions (of the larger intervention study) to document the topographies of challenging behaviors reported by the mother. Each child engaged in a variety of challenging behaviors, including self-injurious behaviors (e.g., head hitting, hand biting); aggression (e.g., biting, hitting, kicking); and other disruptive behaviors (e.g., throwing toys, screaming, mouthing toys).

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