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

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

a University of Oregon, United States  
b University of California-Davis MIND Institute, United States  
c Texas A&M University, United States

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A B S T R A C T

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, & Hessl, 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, & Hessl, 2012). Such...
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, Byers, 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; Sloaneem, 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).
Table 1

Participant characteristics.

<table>
<thead>
<tr>
<th>Participant</th>
<th>Age</th>
<th>PLS-5</th>
<th>Mullen TLAE</th>
<th>Receptive LAE</th>
<th>Expressive LAE</th>
<th>ADOS (Module 1) severity score</th>
<th>Topographies of challenging behaviors</th>
</tr>
</thead>
<tbody>
<tr>
<td>Alex</td>
<td>39</td>
<td>17</td>
<td>20</td>
<td>10</td>
<td>4-ASD</td>
<td>SIB: hitting head</td>
<td>Aggressive behaviors: hitting</td>
</tr>
<tr>
<td></td>
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<td></td>
<td></td>
<td></td>
<td>Disruptive behaviors: elopement</td>
<td></td>
</tr>
<tr>
<td>Bradley</td>
<td>35</td>
<td>26</td>
<td>27</td>
<td>22</td>
<td>4-ASD</td>
<td>SIB: hitting head</td>
<td>Aggressive behaviors: hitting</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Disruptive behaviors: hitting</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Disruptive behaviors: elopement, mouthing objects</td>
<td>Disruptive behaviors: elopement, throwing objects</td>
</tr>
<tr>
<td>Curtis</td>
<td>40</td>
<td>12</td>
<td>11</td>
<td>8</td>
<td>6-AUT</td>
<td>SIB: hitting head</td>
<td>Disruptive behaviors: elopement</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Disruptive behaviors: moaning, throwing objects</td>
<td>Disruptive behaviors: elopement, throwing objects</td>
</tr>
<tr>
<td>David</td>
<td>27</td>
<td>17</td>
<td>10</td>
<td>9</td>
<td>N/A</td>
<td>SIB: hitting head</td>
<td>Disruptive behaviors: hitting</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Disruptive behaviors: crying, throwing objects</td>
<td>Disruptive behaviors: elopement, throwing objects</td>
</tr>
<tr>
<td>Ernest</td>
<td>40</td>
<td>20</td>
<td>24</td>
<td>10</td>
<td>4-ASD</td>
<td>SIB: head banging</td>
<td>Disruptive behaviors: elopement</td>
</tr>
<tr>
<td></td>
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<td></td>
<td></td>
<td></td>
<td>Disruptive behaviors: crying, throwing objects</td>
<td>Disruptive behaviors: elopement, throwing objects</td>
</tr>
<tr>
<td>Franklin</td>
<td>43</td>
<td>26</td>
<td>27</td>
<td>16</td>
<td>4-ASD</td>
<td>SIB: head banging</td>
<td>Disruptive behaviors: elopement</td>
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<tr>
<td></td>
<td></td>
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<td></td>
<td></td>
<td></td>
<td>Disruptive behaviors: crying, throwing objects</td>
<td>Disruptive behaviors: elopement, throwing objects</td>
</tr>
<tr>
<td>Greg</td>
<td>49</td>
<td>30</td>
<td>33</td>
<td>26</td>
<td>6-AUT</td>
<td>SIB: hitting head</td>
<td>Disruptive behaviors: hitting</td>
</tr>
<tr>
<td></td>
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<td></td>
<td></td>
<td></td>
<td></td>
<td>Disruptive behaviors: crying</td>
<td></td>
</tr>
<tr>
<td>Connor</td>
<td>33</td>
<td>9</td>
<td>10</td>
<td>5</td>
<td>6-AUT</td>
<td>SIB: head banging</td>
<td>Disruptive behaviors: elopement</td>
</tr>
<tr>
<td></td>
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<td></td>
<td></td>
<td></td>
<td></td>
<td>Disruptive behaviors: avoiding, crying</td>
<td>Disruptive behaviors: elopement, throwing objects</td>
</tr>
<tr>
<td>John</td>
<td>46</td>
<td>27</td>
<td>28</td>
<td>22</td>
<td>1-Non-spectrum</td>
<td>SIB: head banging</td>
<td>Disruptive behaviors: hitting</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Disruptive behaviors: kicking</td>
<td></td>
</tr>
<tr>
<td>Leon</td>
<td>33</td>
<td>N/A</td>
<td>N/A</td>
<td>18</td>
<td>N/A</td>
<td>N/A</td>
<td>N/A</td>
</tr>
<tr>
<td>Mike</td>
<td>31</td>
<td>21</td>
<td>23</td>
<td>15</td>
<td>4-AUT</td>
<td>SIB: hitting head</td>
<td>Disruptive behaviors: hitting</td>
</tr>
<tr>
<td></td>
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<td></td>
<td></td>
<td></td>
<td>Disruptive behaviors: kicking</td>
<td></td>
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<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Disruptive behaviors: licking arm, throwing objects</td>
<td>Disruptive behaviors: hitting, hitting</td>
</tr>
<tr>
<td>Robert</td>
<td>51</td>
<td>11</td>
<td>14</td>
<td>5</td>
<td>10-AUT</td>
<td>SIB: hitting head</td>
<td>Disruptive behaviors: hitting</td>
</tr>
<tr>
<td></td>
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<td></td>
<td></td>
<td></td>
<td></td>
<td>Disruptive behaviors: hitting, kicking</td>
<td>Disruptive behaviors: elopement</td>
</tr>
<tr>
<td></td>
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<td></td>
<td></td>
<td></td>
<td></td>
<td>Disruptive behaviors: elopement, throwing objects</td>
<td>Disruptive behaviors: elopement, throwing objects</td>
</tr>
</tbody>
</table>

* Age provided in months.
+ PLS total language age equivalence in months.
- Language age equivalence in months.
$^{d}$ ADOS – Toddler module was used and resulted in no severity score.
$^{e}$ Missing data due to participant not tolerating unfamiliar people in room.

Each child was assessed by an experienced masters-level speech/language clinician using the Mullen Scales of Early Learning (MSEL; Mullen, 1995) and the Preschool Language Scale 5th Edition (PLS-5; Zimmerman, Steiner, & Pond, 2011). A different examiner trained to research reliability standards administered the ADOS-2 (Lord et al., 2012). Child characteristics and topographies of challenging behaviors are presented in Table 1.

2.2. Setting and sessions

All assessment and functional analysis sessions were conducted at a university-based clinic. The clinic room was 4 m × 6 m and included a child-sized table and chairs, several adult-sized chairs, an interlocking foam play mat, developmentally appropriate toys, and several closed storage cabinets. During functional analysis sessions, the child, a researcher serving as a parent coach, and the child’s mother were present. One participant (i.e., Mike) engaged in high rates of socially avoidant behavior (crying, hiding face, attempting to leave room) in the presence of non-familiar adults; therefore the parent coach coached his mother via a Bluetooth headset linked to a smartphone from the adjoining observation room. Prior to beginning this study, all data collectors, reliability observers, and other researchers were trained by the first author to implement the procedures and record data. Parent coaches were trained in their roles in the functional analysis through lecture, discussion, video modeling, and role play with performance feedback until each coach implemented functional analysis conditions during role play with 90% accuracy over three consecutive implementations. Training of data collectors and reliability observers included lecture, discussion, and coding video tapes of other children during functional analysis conditions and continued until interobserver agreement was 90% over five consecutive observations.

Mothers were trained for their role in the functional analysis through handouts and a parent coach led discussion of the rationale and procedures for each condition of the functional analysis before the assessment. Immediately before each condition the parent coach briefly reviewed the procedures with the mother. During each condition, the parent coach provided the least amount of prompting (i.e., gestural prompt, verbal prompt, model prompt) needed to evoke correct implementation of assessment procedures by the mother.
2.3. Experimental design

Individual multielement designs were used to demonstrate experimental control (Gast, 2010). Participants were exposed to five different experimental conditions (i.e., attention, social avoidance, demand, tangible, and play), which were implemented in a randomized fashion by the child's mother using a modified version of the Iwata et al. (1982–1994) methodology as described in Section 2.5.

2.4. Procedure

Mothers of each participant completed the Questions About Behavioral Function (QABF; Matson & Vollmer, 1995) prior to implementation of the functional analysis. Functions were coded for those QABF subscales on which the parent indicated a score of 10 or higher (4 of the 6 items of a subscale needed to be endorsed to obtain a score of 10). Data obtained from the physical discomfort subscales are not reported because we did not assess this putative function within the functional analysis. Similarly, the functional analysis included a social avoidance condition to determine whether challenging behavior was maintained by negative reinforcement in the form of escape from or avoidance of adult attention (cf., Hagopian, Wilson, & Wilder, 2001), although the QABF does not include this category.

2.5. Experimental functional analysis

Functional analyses were implemented over the course of one to two days with a total duration of no more than 45 min each day. Between 7 and 15 (M = 13) 5-min conditions were conducted with each child. The attention condition assessed whether challenging behavior was sensitive to positive reinforcement in the form of maternal attention. In this condition, the child was free to engage with preferred toys, and the mother was instructed not to interact with her child unless the target challenging behavior occurred, at which point she delivered attention for approximately 10 s.

The social avoidance condition assessed whether challenging behavior was sensitive to negative reinforcement in the form of escape from social interaction. During this condition, the mother provided continuous attention to the child (i.e., enthusiastically commenting on play and frequent positive touch) while the child was free to play with a variety of preferred toys (identified by paired choice preference assessment; Lohmann-O’Rourke & Browder, 1998). The mother provided attention unless challenging behavior occurred, in which case she refrained from interacting with her child for approximately 10 s (see Hagopian et al., 2001).

The demand condition assessed whether challenging behavior was maintained by negative reinforcement in the form of escape from task demands. The child was engaged in a variety of pre-academic instructional activities (e.g., putting together a puzzle, sorting shapes stringing beads). Contingent upon challenging behavior, the mother removed all materials for a minimum of 10 s and then immediately re-introduced materials once the challenging behavior had ceased.

The tangible condition assessed whether challenging behavior was sensitive to positive reinforcement in the form of access to preferred tangibles. The child was given brief access (30-s) to a highly preferred toy (with the most highly preferred toy identified by paired choice preference assessment; Lohmann-O’Rourke & Browder, 1998). Following this period of brief access, the mother held the toy in sight, but out of reach of the child. Contingent on the challenging behavior, the mother delivered the preferred toy for approximately 10 s.

The play condition served as a control condition as demands were absent and maternal attention was freely available. During the play condition, the child was free to engage in a variety of activities (e.g., playing with favorite toys) that they were observed to enjoy during pre-assessment observations. Irrespective of the child's behavior during play, the mother interacted pleasantly with her child approximately every 10 s.

2.6. Data collection and analysis

Target challenging behaviors (as reported by mothers during the pre-assessment interview) were individually coded across all functional analysis sessions using a 10 s partial interval recording procedure (Gast, 2010). The percentage of 10 s intervals in which the target challenging behavior occurred was calculated for each 5-min session. Data were collected via a one-way observation window and digitized video-recording equipment.

The percentages of 10 s intervals with target challenging behaviors during each of the assessment observations were plotted on a line graph. To determine if individual topographies of challenging behavior were exclusive to any one functional analysis condition, topographies were graphed both in aggregate and separately (Derby et al., 1994, 2000). To supplement visual analysis, a modified version (see Martin, Gaffan, & Wiliams, 1999) of the Hagopian et al. (1997) criterion for differentiation across social conditions was used to determine the social consequences maintaining challenging behavior. The modified method requires calculation of the mean of the control condition (i.e., play) and drawing a criterion line (CL) 1 standard deviation above the mean on the line graph. At least one half of the data points for any condition must lie above the CL to assign a behavioral function. When the occurrence of target challenging behavior in the play condition was near zero, the CL was set at 8.33% of intervals (i.e., minimum of .5 responses per min; Langthorne et al., 2011). Analysis of separately graphed topographies of challenging behavior demonstrated that individual topographies were not exclusive to any one functional analysis condition.
Table 2
Interobserver agreement data for challenging behavior data for participants 1–12.

<table>
<thead>
<tr>
<th>Participant</th>
<th>Mean percentage and range of agreement</th>
</tr>
</thead>
<tbody>
<tr>
<td>Alex</td>
<td>99% (97–100%)</td>
</tr>
<tr>
<td>Bradley</td>
<td>99% (97–100%)</td>
</tr>
<tr>
<td>Curtis</td>
<td>92% (77–100%)</td>
</tr>
<tr>
<td>David</td>
<td>92% (83–100%)</td>
</tr>
<tr>
<td>Ernest</td>
<td>92% (77–100%)</td>
</tr>
<tr>
<td>Franklin</td>
<td>94% (83–90%)</td>
</tr>
<tr>
<td>Greg</td>
<td>89% (83–93%)</td>
</tr>
<tr>
<td>Connor</td>
<td>87% (77–100%)</td>
</tr>
<tr>
<td>John</td>
<td>95% (90–97%)</td>
</tr>
<tr>
<td>Leon</td>
<td>81% (80–82%)</td>
</tr>
<tr>
<td>Mike</td>
<td>89% (80–89%)</td>
</tr>
<tr>
<td>Robert</td>
<td>90% (87–97%)</td>
</tr>
</tbody>
</table>

2.7. Interobserver agreement and procedural fidelity

A second observer independently coded challenging behavior data during 33% (Alex, Ernest, Franklin, Conner), 29% (Leon), and 20% (Bradley, Curtis, David, Greg, John, Mike, Robert) of sessions. Interobserver agreement for each 5 min session was calculated using the total agreement procedure (i.e., dividing the total number of agreements by the total number of intervals in the session and multiplying by 100; Gast, 2010). Overall mean interobserver reliability ranged from 81% to 99% with a mean of 92%. Interobserver agreement data for challenging behavior is presented for each child in Table 2.

Fidelity of caregiver implementation of functional analysis procedures was assessed on 100% of sessions for each child. Task analyses of anticipated caregiver behaviors for conditions were used to assess the number of steps completed correctly by each parent. Each step was recorded as correct (+) if the step was completed and incorrect (−) if the step was omitted, not completed correctly, or was completed out of sequential order. The percentage of steps completed correctly was calculated for each implemented condition by dividing the number of steps completed correctly by the total number of steps and multiplying by 100 to obtain a percentage. Overall mean procedural fidelity ranged from 85% to 93% with a mean of 89%.

3. Results

Fig. 1 presents a summary of functional analysis results for Alex (panel 1), Bradley (panel 2), Curtis (panel 3), David (panel 4), Ernest (panel 5), and Franklin (panel 6). Fig. 2 presents a summary of functional analysis results for Greg (panel 1), Connor (panel 2), John (panel 3), Leon (panel 4), Mike (panel 5), and Robert (panel 6). With the exception of Curtis (tangible only), Franklin (escape only), Connor (inconclusive), Leon (tangible), and Mike (tangible only) challenging behavior occurred at Hagopian et al.’s (1997) criterion levels for each participant across multiple functional analysis conditions.

Alex engaged in relatively higher rates of challenging behavior in the social avoidance (M = 6.67%, range = 0–10%) and attention (M = 6.65%, range = 3.3–10%) conditions as compared to the other conditions. He engaged in low or zero levels of challenging behavior in the demand (M = 4.33%, range = 3–6.7%) and play (M = 1%, range = 0–3%) conditions. There were variable but overall lower levels of challenging behavior in the tangible (M = 5.33%, range = 0–16%) condition.

Bradley engaged in high rates of challenging behavior in the tangible (M = 63.5%, range = 63–64%) and demand (M = 47, range = 37–57%) conditions and in one half of the attention (M = 45%, range = 3–87%) condition sessions. There were low or zero levels of challenging behavior during both the play (M = 3.5%, range = 0–7%) and social avoidance (M = 0%) conditions.

Curtis engaged in variable, but relatively higher levels of challenging behavior during the tangible condition (M = 12.33%, range = 7–17%) than in the other conditions and generally low levels of challenging behavior during the demand (M = 5.67%, range = 0–13%) condition. There were zero levels of challenging behavior during the play (M = 0%), social avoidance (M = 0%), and attention (M = 0%) conditions.

David engaged in higher rates of challenging behavior in the demand (M = 34.33%, range = 23–43%) and tangible (M = 25.67%, range = 17–40%) conditions. He engaged in low or zero levels of challenging behavior in the play (M = 0%), attention (M = 2%, range = 0–3%), and social avoidance (M = 0%) conditions.

Ernest engaged in the highest rates of challenging behavior during the demand (M = 59%, range = 0–97%) and tangible (M = 32%, range = 23–40%) conditions than in the other conditions. He engaged in variable, but generally low rates of challenging behavior during the social avoidance (M = 6.67, range = 0–20%) and attention (M = 14.56, range = 0–37%) conditions. He engaged in zero rates of challenging behavior during the play (M = 0%) condition.

Franklin engaged in the highest rates of challenging behavior during the demand (M = 33.33, range = 0–53.33%) and tangible (M = 10%, range = 13.33–20%) conditions and variable, but generally low rates of challenging behavior during the play (M = 11.11, range = 3.33–23.33%), attention (M = 3.33, range = 0–10), and social avoidance (M = 5.56, range = 0–10%) conditions.

Greg engaged in higher rates of challenging behavior during the demand (M = 63.33, range = 50–83%), tangible (M = 17.67, range = 3–33%) and attention (M = 15.67, range = 0–30%) conditions than in the other conditions, and low or
zero rates of challenging behavior during the play ($M = 2\%$, range $= 0–3\%$) and social avoidance ($M = 3.33\%$, range $= 0–7\%$) conditions.

Connor engaged in the highest rates of challenging behavior during the demand ($M = 17.89\%$, range $= 7–36.67\%$) and tangible ($M = 10\%$, range $= 10–10\%$) conditions, but variable and generally low rates of challenging behavior during the attention ($M = 2.22\%$, range $= 0–6.67\%$), play ($M = 26.67\%$, range $= 0–20\%$), and social avoidance ($M = 1\%$, range $= 0–3\%$) conditions.

John engaged in the highest rates of challenging behavior during the demand ($M = 11.5\%$, range $= 10–13\%$), tangible ($M = 8.5\%$, range $= 7–10\%$), and social avoidance ($M = 10\%$, range $= 7–13\%$) conditions. He engaged in much lower rates of challenging behavior during the play ($M = 1\%$, range $= 0–3\%$) and attention ($M = 3.5\%$, range $= 0–7\%$) conditions.

Leon engaged in higher rates of challenging behaviors during the tangible (80\%), demand (33\%), and play (22.11\%, range $= 3.33–40\%$) conditions than in the other conditions. He engaged in lower levels of challenging behavior during the social avoidance (7\%) and attention (10\%) conditions. Mike engaged in increased rates of challenging behaviors during the demand ($M = 38.89\%$, range $= 3.33–100\%$) and tangible (53.33\%) conditions. He engaged in generally low levels of challenging behaviors during the attention ($M = 1.11\%$, range $= 0–3.33\%$), play ($M = 6.67\%$, range $= 0–20\%$), and social avoidance ($M = 11.11\%$, range $= 3.33–23.33\%$) conditions.

Robert engaged in the highest rates of challenging behaviors during the demand ($M = 14.44\%$, range $= 0–23.33\%$), tangible ($M = 23.33\%$, range $= 13.33–33.33\%$), and social avoidance ($M = 16.67\%$, range $= 3.33–36.67\%$) conditions. He engaged in lower levels of challenging behavior during the play ($M = 2.22\%$, range $= 0–6.67\%$) and attention ($M = 12.22\%$, range $= 0–33.33\%$) conditions.

A summary of the results of the QABF and the functional analyses for each child are presented in Table 3. Complete correspondence between the results of the QABF and the functional analysis was obtained for 3 of the 12 children (i.e., Bradley, David, and Robert). Partial correspondence (i.e., the QABF and functional analysis indicated agreement on at least one social function of challenging behavior) was obtained for 5 of the remaining 8 children (i.e., Alex, Curtis, Ernest, Greg, and John). QABF and functional analysis results did not agree for 4 of the children (i.e., Franklin, Connor, Leon, or Mike).
Fig. 2. Percentage of intervals with challenging behavior across tangible, demand, attention, social avoidance, and play conditions for Greg (panel 1), Connor (panel 2), John (panel 3), Leon (panel 4), Mike (panel 5), and Robert (panel 6).

Table 3
Operant functions identified by Questions About Behavioral Function (QABF) and Functional Analysis.

<table>
<thead>
<tr>
<th>Participant</th>
<th>QABF</th>
<th>Functional analysis</th>
</tr>
</thead>
<tbody>
<tr>
<td>Alex</td>
<td>Escape</td>
<td>Attention, Escape (SA)</td>
</tr>
<tr>
<td>Bradley</td>
<td>Attention, Escape, Tangible</td>
<td>Attention, Escape (demand), Tangible</td>
</tr>
<tr>
<td>Curtis</td>
<td>Attention, Escape, Tangible</td>
<td>Escape (demand), Tangible</td>
</tr>
<tr>
<td>David</td>
<td>Escape, Tangible</td>
<td>Escape (demand), Tangible</td>
</tr>
<tr>
<td>Ernest</td>
<td>Escape</td>
<td>Escape (demand), Tangible</td>
</tr>
<tr>
<td>Franklin</td>
<td>Tangible</td>
<td>Escape (demand)</td>
</tr>
<tr>
<td>Greg</td>
<td>Escape, Tangible</td>
<td>Attention, Escape (demand), Tangible</td>
</tr>
<tr>
<td>Connor</td>
<td>Escape, Tangible</td>
<td>Inconclusive</td>
</tr>
<tr>
<td>John</td>
<td>Attention, Escape, Tangible</td>
<td>Escape (demand and SA), Tangible</td>
</tr>
<tr>
<td>Leon</td>
<td>Escape</td>
<td>Tangible</td>
</tr>
<tr>
<td>Mike</td>
<td>Attention, Escape</td>
<td>Tangible</td>
</tr>
<tr>
<td>Robert</td>
<td>Escape, Tangible</td>
<td>Escape (demand and SA), Tangible</td>
</tr>
</tbody>
</table>

* Social avoidance.
4. Discussion

In the present study, we implemented functional analyses with 12 young males with FXS. Nine of the 12 children (i.e., Bradley, Curtis, David, Ernest, Greg, John, Leon, Mike, and Robert) engaged in challenging behavior that met Hagopian et al.’s (1997) criteria for tangibly-maintained challenging behavior. Seven children (i.e., Bradley, David, Ernest, Franklin, Greg, John, and Robert) engaged in challenging behavior that met criteria for escape-maintained (demand) behavior. Three children (i.e., Alex, John, and Robert) engaged in challenging behavior that met criteria for escape-maintained (social avoidance) behavior. Connor’s functional analysis results were inconclusive. That is, although he engaged in challenging behaviors in all functional analysis conditions, Connor did not engage in the necessary frequency of challenging behavior to meet criteria for any operant function of challenging behavior. Three children (i.e., Alex, Bradley, Greg) engaged in levels of challenging behavior that met criteria for attention-maintained challenging behavior. Our findings have established the early emergence of challenging behaviors in a more experimentally rigorous method than previous studies. In the current study, the majority of children engaged in tangible and escape-maintained (demand) challenging behavior and fewer engaged in attention maintained challenging behavior. These findings corroborate past research reporting the results of indirect (i.e., informant report) functional behavior assessments (Symons et al., 2003; Woodcock et al., 2009) and experimental functional analyses of challenging behavior with older individuals with FXS (Langthorne et al., 2011).

Similar to Langthorne et al. (2011) findings, only three children in the present study engaged in challenging behavior to escape social interactions with their mother during the social avoidance condition of the functional analysis. These findings would seem to support Langthorne et al.’s (2011) hypothesis that adult attention does not function as an aversive stimulus. The majority of children in the present study engaged in low or zero levels of challenging behavior during attention conditions and thus, did not contact the condition’s arranged contingencies. However, we observed that many of the children engaged in appropriate communicative behaviors (e.g., smiling in direction of mother, standing close to mother, vocalization) in an apparent attempt to obtain their mother’s attention during the attention conditions. In accordance with functional analysis methodology, appropriate communicative behaviors were not reinforced during the attention condition; however, these observations suggest that young children with FXS value their mother’s attention, but do not engage in challenging behavior to obtain her attention. Future research should continue to experimentally evaluate the role of adult attention in the maintenance of challenging behavior for children with FXS. Researchers should also evaluate the possible influence of autism symptomology on the attention-seeking behaviors of children with FXS. Past research has shown that attention-maintained challenging behaviors are common for children with autism spectrum disorder (e.g., Love, Carr, & LeBlanc, 2009). We were unable to relate our observations of pro-social attention getting behaviors during functional analyses to participants’ ADOS scores because we did not take empirical data on appropriate behavior.

Our results confirm previous empirical findings indicating that challenging behaviors such as self-injurious behaviors often appear during early childhood in males with full mutation FXS (Bailey et al., 2008; Symons et al., 2010). Symons et al. (2010) found that the most commonly reported onset of self-injurious behavior for males was between 1 and 3 years of age and the most common topographies of self-injurious behavior for males were biting/chewing self (69.6% of respondents) typically directed toward hands or arms and hitting self (39.4% of respondents). According to parent report, one half of the children in the present study engaged in head bumping or head hitting and 25% of the children engaged in hand biting. An additional two children engaged in hand mouthing or arm licking. Our use of functional analysis methodology allowed us to observe and confirm maternal reports of self-injurious, aggressive, and other disruptive behaviors. Future research should emphasize the evaluation of function-based behavioral interventions for children with FXS during early childhood to decrease or prevent the worsening of self-injurious and aggressive behaviors. Demonstrating the effectiveness of interventions based on functional analysis results would also validate the functional analysis results.

The functional analysis procedures of the current study differed from those used by Langthorne et al. (2011) and Lesniak-Karpiak et al. (2003) in that the child’s biological mother implemented all sessions. Past studies including individuals with FXS have generally used experimenters to implement social escape and social interaction conditions (Langthorne et al., 2011; Lesniak-Karpiak et al., 2003) and have found low levels of challenging behavior during assessment conditions. Our findings are notably similar to these studies. That is, the majority of children in our study did not engage in criterion levels of challenging behavior during attention or social avoidance conditions. A unique contribution of the current study is the use of biological mothers, rather than experimenters, to implement functional analysis procedures. These procedures improve the ecological validity of our findings. Cohen et al. (1988) compared social escape behaviors during experimenter and parent (i.e., mother) implemented social situations and found that children with FXS and Down syndrome were more likely to engage in escape behaviors when interacting with a non-familiar adult, whereas children with diagnoses of autism and atypical pervasive developmental disorder did not discriminate between their parent and a stranger. However, children with FXS were more likely to avoid both parent and stranger than children with Down syndrome. These mixed results suggest the need for additional research comparing the results of social avoidance and attention conditions when implemented by familiar and non-familiar adults. In the context of the parent-implemented language intervention study for which the current sample was recruited, it was deemed important to evaluate the presence of challenging behavior with the mother as she would be acting as the primary interventionist for her child.

There are several limitations to the present study that necessitate future research. First, the majority of children in the current study displayed behaviors frequent and severe enough to warrant ADOS-2 (Lord et al., 2012) scores indicative of ASD. In the only other study using functional analysis methodology, only one of eight participants was reported to have a
diagnosis of autism, but ADOS scores were not reported (Langthorne et al., 2011). In analyzing data from the National Fragile X Survey, Symons et al. (2010) identified differences in the presence of co-existing conditions (e.g., autism, anxiety, depression) for 258 pairs of boys with FXS who were matched according to the presence or absence of self-injurious behaviors. Boys for whom self-injurious behavior was reported during the 30 days prior to completion of the survey were significantly more likely to have a diagnosis of autism than boys for whom self-injurious behavior was not reported. Thus, the rates of challenging behavior we observed across conditions of the functional analysis may in part have covaried with our participants’ autism severity scores. Alternatively, a negative association is observed in FXS between autism symptomology and nonverbal cognitive ability (Lewis et al., 2006), a factor also implicated as a risk factor in the presence of challenging behaviors (Cinic, Hoffman, Gaze, & Edelbrock, 2004). More research is warranted to understand how child characteristics influence the development of challenging behaviors in FXS.

Additionally, our participants were younger than those included in past studies and it is possible that other young children matched to the language and cognitive abilities of our participants might have engaged in similar challenging behaviors during functional analysis conditions. To determine whether the behavioral patterns we observed are unique to children with FXS, future research examining the operant function of challenging behavior should endeavor to include comparison groups of children, such as children with idiopathic ASD and children with FXS who are matched on such characteristics as autism severity scores, nonverbal cognition, and language ability. The ADOS-2 could be used to quantify symptoms of ASD using calibrated severity scores (Gotham, Pickles, & Lord, 2009). The use of autism severity scores that are computed separately for the social affective and repetitive behavior domains (Hus, Gotham, & Lord, 2013) may also help to clarify which symptoms of ASD covary with challenging behaviors in young boys with FXS.

Second, our lack of an alone condition in the functional analysis to assess the presence of automatically rather than multiply maintained challenging behavior does not allow us to rule out automatically maintained challenging behavior. The young age of the children in the present study prevented the implementation of alone conditions due to the developmentally appropriate distress experienced by young children when left alone in an unfamiliar place. Langthorne et al. (2011) implemented a no-interaction condition and found that no participant engaged in criterion levels of challenging behavior during these conditions. Langthorne et al. pointed out that a no-interaction condition may not be the most accurate condition to assess automatically maintained challenging behavior, because salient discriminative stimuli (i.e., experimenter physically present) for the delivery of attention are available, and the condition starts with deprivation of attention, a putative establishing operation (EO) for attention-maintained challenging behavior. High rates of challenging behavior during no interaction conditions could indicate attention-maintained challenging behavior rather than automatically maintained challenging behavior (O’Reilly et al., 2006). Nevertheless, inclusion of this condition would allow assessment of child behavior in the absence of adult interaction. Future research with young children should include no-interaction conditions during functional analysis or assess the rate of challenging behavior during naturally occurring times when the child’s caregiver is not present (e.g., child plays in bedroom with caregiver in adjacent room).

Third, we obtained mixed correspondence between the QABF (Matson & Vollmer, 1995) and functional analysis results with complete correspondence found for only 3 of the 12 children. Recent research has demonstrated good correspondence between these assessments for children with ASD (Healy, Brett, & Leader, 2013; Watkins & Rapp, 2013). However, our mixed findings are in agreement with those of other researchers comparing indirect functional behavior assessment results to experimental functional analysis results for older individuals with FXS (Langthorne et al., 2011). Despite congruence with the results of past research with this population, examination of the comparative effectiveness of function-based interventions derived from indirect functional behavior assessments such as the QABF and functional analysis procedures may better inform researchers and practitioners of the clinical utility of indirect functional behavior assessments in identifying putative social and non-social consequences maintaining challenging behaviors for individuals with FXS.

Due to scheduling constraints and child fatigue, it was not always possible to continue functional analysis until we achieved stable responding across a condition. For instance, Leon’s functional analysis consisted of 3 observations of the play condition, and 1 observation each of the attention, tangible, demand, and social avoidance conditions. Additionally, Connor’s functional analyses results were inconclusive. However, the efficacy of brief functional analysis procedures (i.e., those with two or fewer observations per condition) when compared to full functional analysis procedures (i.e., those with three or more observations for two or more condition) has been demonstrated (Derby et al., 1992; Hanley et al., 2003; Kahng & Iwata, 1999; Northup et al., 1991). Brief functional analysis procedures are often used in outpatient clinics where logistical constraints necessitate a shortened assessment (Wacker et al., 1994), but the ability of our findings to extend our understanding of the behavioral phenotype of FXS would be strengthened by extended functional analysis procedures (Iwata et al., 1982–1994) conducted with a larger sample of children until stable responding was obtained across conditions.

5. Conclusions

Despite the aforementioned limitations, this is the first study to report the operant function of challenging behavior for young children with FXS (27–51 months). The current findings, and those reported by other researchers (e.g., Langthorne et al., 2011), suggest a pattern of challenging behavior in persons with FXS that may be difficult to effectively address. For instance, communication intervention is a priority for young children with FXS who present with language delays, but successful implementation may be hindered by a child’s challenging behavior. Utilizing experimental functional analysis procedures to guide behavioral intervention may be essential to assisting interventionists and parents to effectively address
challenging behavior that interferes with daily routines and activities. Moreover, intervening early to prevent the worsening of existent challenging behavior may yield improved distal outcomes for children with FXS and their families.

Conflict of interest

The authors declare that they have no conflict of interest.

References


