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POSITIVE FAMILY INTERVENTION FOR FAMILIES OF CHILDREN WITH
FRAGILE X SYNDROME: USING TELEHEALTH TO REDUCE PROBLEM
BEHAVIOR AND IMPROVE MATERNAL MENTAL HEALTH

A dissertation submitted in partial fulfillment
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New York

by

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ABSTRACT

POSITIVE FAMILY INTERVENTION FOR FAMILIES OF CHILDREN WITH FRAGILE X SYNDROME: USING TELEHEALTH TO REDUCE PROBLEM BEHAVIOR AND IMPROVE MATERNAL MENTAL HEALTH

Carmen Ann Kemper Mootz

The present study was designed to evaluate the effectiveness of Positive Family Intervention (PFI), delivered via teletherapy, for a family of a child diagnosed with Fragile X syndrome (FXS). PFI combines elements of cognitive-behavioral therapy (CBT) and positive-behavior support (PBS). A single-subject AB design was used across one participant to evaluate changes in observed problem behavior. Following nine weeks of baseline data collection, the participating mother was administered PFI via WebEx for eight 90-minute sessions, once per week. After the treatment was complete, three weeks of post-treatment follow-up data on problem behavior were collected from video-recordings. The participant uploaded naturalistic videos of her child's problem behavior in a specific setting to a secure online server. The effect of PFI on parental depression, stress, and cognitions was also evaluated using pre- and post- treatment data collection. Results indicated a substantial decrease in both observed and parent-reported problem behavior over the course of the treatment. Non-significant decreases were reported in parental depression and stress. Small to medium changes in parental attribution of child problem behavior were also reported following treatment. The implications of these changes are discussed, as well as the limitations of the present study and directions for future research.

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INTRODUCTION

Behavioral interventions, particularly behavioral parent training programs, have demonstrated effectiveness for reducing problem behaviors in children with autism spectrum disorder (ASD) and other developmental disabilities (DD) (Brosnan & Healy, 2011; Heyvaert, Saenen, Campbell, Maes, & Onghena, 2014; Matson, Mahan, & LoVullo, 2009; Petrenko, 2013; Wong et al., 2015). Given the effectiveness of such interventions for those with ASD and DD, it is surprising that relatively little research has been conducted on the effectiveness of parent training interventions to ameliorate problem behaviors in children with Fragile X syndrome (FXS; see Moskowitz & Jones, 2015 for a review of the limited behavioral intervention research in FXS).

FXS is the most common inherited cause of intellectual disability, and the behavioral phenotype of the disorder includes numerous problem behaviors such as aggression and self-injury (Hall, Lightbody, & Reiss, 2008). In fact, behavioral challenges are one of the most common concerns of families who have a child with FXS (Wheeler et al, 2014). Yet FXS has been largely absent from research on behavioral interventions generally, including studies on parent training to address child problem behavior. In addition to a lack of research on behavioral interventions, there is also a lack of behavior management services available to children with FXS who might benefit from them (Martin et al., 2013). Access to evidence-based treatment is a barrier to many families of children with DD (Elder, Brasher, & Alexander, 2016; Patel, Kieling, Maulik, & Divan, 2013; Pickard, Kilgore, & Ingersoll, 2016). This is likely to be especially true for families of children with FXS, given that FXS is a rare disorder. With individuals with FXS scattered across the country, alternative modes of delivery for evidence-based

treatment, such as computer-delivered parent training (telehealth), must be evaluated in order to determine if they are an effective alternative to in-person treatment for parents of children with FXS. Research additionally indicates that parental functioning is associated with child problem behaviors in FXS; mothers with high levels of depression and stress report higher levels of problem behavior in their children with FXS (Wheeler, Hatton, Reichardt & Bailey, 2007). This suggests that there is a need for treatments that target parents' thoughts and emotions in the context of parent training for parents of youth with FXS. A multisite randomized clinical trial demonstrated that Positive Family Intervention (PFI), a manualized behavioral parent training program that integrates Positive Behavior Support (PBS) with Cognitive Behavioral Therapy (CBT), resulted in improvements in child problem behavior and parental pessimism for families of children with other DDs (not including FXS). The present project aims to examine whether PFI delivered via distance teleconferencing would result in similar improvements in child problem behavior as well as parental functioning for a family of a child with FXS.

Fragile X Syndrome

Fragile X syndrome (FXS) is the most common known inherited cause of intellectual disability (ID) (Mila, Alvarez-Mora, Madrigal, & Rodriguez-Revenga, 2017) and the leading known genetic cause of autism (Reddy, 2005). FXS is a genetic disorder brought about by a mutation in the FMR1 gene, causing impaired production of the Fragile X mental retardation protein (FMRP) due to repeating patterns of CGG DNA (Saldarriaga, Tassone, González-Teshima, Forero-Forero, Ayala-Zapata, & Hagerman, 2014). Individuals without FXS typically have 45 CGG repetitions on the FMR1 gene; those with full-mutation FXS will have upwards of 200 repeats (Mila et al., 2017). This leads to increased methylation, resulting in a lack of FMRP. While both males and

females can inherit FXS, it is more common in males (roughly 1.4 per 10,000 males and 0.9 per 10,000 females; Hunter, Rivero-Arias, Angelov, Kim, Fotheringham, & Leal, 2014) due to the FMR1 gene's location on the X chromosome. The location of the FMR1 gene also contributes to the typically more severe presentation of FXS in males (Roberts, Weisenfeld, Hatton, Heath, & Kaufmann, 2007). The majority of males who have FXS and no other comorbid diagnoses are likely to have anywhere from mild intellectual disability, with full-scale IQ scores of around 55 to 70 (Boyle & Kaufmann, 2010), to severe intellectual disability, with full-scale IQ scores between 40 to 55 (Berry-Kravis et al., 2016). Around one-third to one-quarter of females with FXS have significant intellectual disability (Coffee et al., 2009).

Behavioral Phenotype of Fragile X Syndrome

Beyond intellectual disability, individuals with FXS present with a distinct behavioral phenotype consisting of impaired eye-contact, hyperarousal, extreme shyness, anxiety, hand biting or hand flapping, perseverative speech, and an increased risk for aggressive behaviors (Boyle & Kaufmann, 2010; Hagerman, 2002; Hagerman et al., 2009). Males with FXS also tend to have impaired verbal skills, particularly in expressive language, and impairments in executive functioning and attention (Boyle & Kaufmann, 2010; Smith, Barker, Seltzer, Abbeduto, & Greenberg, 2012). The FXS phenotype has features similar to other disorders, in particular autism spectrum disorder (ASD) and Attention-Deficit/Hyperactivity Disorder (ADHD); in fact, ASD and ADHD are both frequent comorbid diagnoses in individuals with FXS (Wolff et al., 2012). Even when symptoms fall short of meeting diagnostic criteria, boys with FXS regularly display features of ADHD, anxiety, and ASD (Leitner, 2014); roughly 84% of males with FXS

have also been diagnosed with or treated for attentional problems, 66% for hyperactivity, and 70% for anxiety (Bailey, Sideris, Roberts, & Hatton, 2008).

Problem Behavior in Fragile X Syndrome

Parents and professionals typically report that problem behavior is their greatest concern regarding their children with FXS (Hatton, Hooper, Bailey, Skinner, Sullivan, & Wheeler, 2002), with aggression, self-injurious behavior (SIB), and stereotypic behaviors reported to be the most prevalent externalizing behaviors in FXS. Aggression was reported to occur in over 90% of individuals with FXS (Wheeler, Raspa, Bailey, Bishop, & Bailey, 2016) while SIB was reported to occur in approximately 58-71% of boys with FXS. As many as 91-98% of boys with FXS exhibit at least one stereotypy (Hall et al., 2016; Hessel et al., 2008). Although not as prevalent as aggression, SIB, or stereotypy, tantrums or temper outbursts are also reported by caregivers in about a quarter of children with FXS (Newman, Leader, Chen, & Mannion, 2015; Woodcock, Oliver, & Humphreys, 2009).

The intervention literature for problem behavior in individuals with developmental disabilities (DD) stresses the importance of understanding the function or purpose of an individual's problem behavior before targeting it for change. A meta-analysis by Carr et al. (1999) found that interventions targeting problem behavior in children with DD are about twice as likely to be successful if they are based on assessment of the function of problem behavior. This is because the same behavior may be serving a different function in different children, or may be serving a different function in different situations within the same child. For example, one child may tantrum in class because the teacher's attention is reinforcing, whereas a different child

may tantrum in class in order to escape or avoid social interaction (Taylor & Carr, 1992); these different functions would require different interventions.

Although some research has been done on the topography or types of problem behaviors displayed by children with FXS (Hardiman & McGill, 2016), less is known about the functions of said behavior. Conducting a functional behavior assessment (FBA) involves identifying the antecedents (A) and consequences (C) that maintain problem behavior (B) using indirect measures (questionnaires, interviews), direct observation, and/or an experimental functional analysis. Hall, DeBernardis, and Reiss (2006) found that children with FXS were more likely to display problem behavior in social demand situations than in a non-social task, suggesting the function of their problem behaviors was likely to escape the social and performance demands. Using an indirect assessment (caregiver-report), Langthorne and McGill (2012) determined that children with FXS were less likely to engage in attention-maintained problem behavior compared to children with Smith-Magenis syndrome or other non-specific intellectual and developmental disabilities. Three studies that conducted experimental functional analyses (FAs) with children with FXS (Kurtz et al., 2015; Langthorne et al., 2011; Machalicek et al., 2014) suggest that certain functions – namely, escape and tangible – might be more common in children with FXS than in those with other DDs, whereas the attention function may be less common in FXS than in those with other DDs. These studies highlight the importance of understanding the function of problem behaviors for different populations in order to successfully intervene, although an individualized FBA and intervention plan are always necessary, given that – in spite of a common behavioral phenotype –

individuals with FXS still show substantial within-syndrome variability (Moskowitz & Jones, 2015).

Interventions for Problem Behaviors in Fragile X and Developmental Disorders

Applied behavior analysis (ABA) has been the treatment of choice for children with neurodevelopmental disorders for decades (Matson et al., 2009; Matson, Turygin, Beighley, Rieske, Tureck, & Matson, 2012). ABA uses the principles of operant conditioning to alter behavior (e.g., Dixon, Vogel, & Tarbox, 2012). Matson and colleagues (2009) argued that evidence-based parent training programs should in fact be the “center piece” for interventions geared towards children with intellectual disabilities because of the intense amount of labor involved in these interventions and because of the importance of generalization to multiple contexts. Parent training programs vary, but effective programs typically use behavioral principles to change the pattern of reinforcement in parent-child interactions, thus altering children’s behavior (e.g. Fettig & Barton, 2013; Matson et al., 2009; Michelson et al., 2013). The majority of the research on these parent training programs has involved typically developing children with conduct problems, defiance, or oppositionality, and children with ASD (Aman et al., 2009; Brookman-Frazee, Stahmer, Baker-Ericzen, & Tsai, 2006; Farmer et al., 2012; Postorino et al., 2017).

A substantial body of research demonstrates that family-based Positive Behavior Support (PBS) is an effective intervention for improving behavior problems in children with ASD and other DDs (e.g., Buschbacher, Fox, & Clarke, 2004; Clarke, Dunlap, & Vaughn, 1999; Clarke et al., 2002; Dunlap et al., 2010; Lucyshyn et al., 2007, 2015). PBS is rooted in ABA (Dunlap, Carr, Horner, Zarcone, & Schwartz, 2008) but also

incorporates multiple theoretical perspectives into its framework (Carr et al., 2002). PBS focuses on understanding the function of challenging behaviors and designing multicomponent intervention plans, based on the function, that prevent and manage problem behavior and teach skills to replace the problem behavior (Carr et al., 2002; Dunlap et al., 2008). The model emphasizes person-centered values (e.g., family goals and values), ecological validity, and collaboratively developing interventions with stakeholders and implementing these interventions in naturalistic environments to promote generalization and maintenance (Carr et al., 2002; Lucyshyn et al., 2007), ensuring behavioral changes maintain not only in highly controlled settings but also in homes, schools, and the wider community.

Positive Family Intervention

Positive Family Intervention (PFI) is a manualized parent-training program that integrates PBS (which helps parents to identify and change patterns in their child's behavior) with aspects of cognitive-behavioral therapy (CBT, which helps parents to identify and change their own thoughts and feelings about their children and themselves) (Durand & Hieneman, 2008). In terms of PBS, PFI teaches parents to develop an individualized behavior support plan, based on the functional behavior assessment, that includes intervention strategies to (a) prevent the problem behavior from occurring, (b) manage consequences (minimizing reinforcement for problem behavior while reinforcing positive behavior), and (c) teach the child more appropriate skills to replace the problem behavior.

In terms of CBT, PFI teaches parents to assess and intervene on their own thoughts, which has the potential to improve parental functioning. Durand and colleagues

(2013) found that parental pessimism decreased after completing PFI. The improvement of parental mental health is an important but regularly overlooked aspect of treating child problem behaviors. A large body of evidence indicates that poor parental mental health is a major factor in treatment outcome for individuals with ASD/DD (Abbeduto et al., 2004; Durand et al., 2013; Whittingham, Sofronoff, Sheffield, & Sanders, 2008) and FXS (Abbeduto et al., 2004; Hauser, Kover, and Abbeduto, 2014). Poor parental functioning can increase rates of dropout, negatively influence treatment fidelity, and interfere with maintenance of treatment gains (Hauser et al, 2014).

The increased demands of parenting a child with a developmental disability have been found to negatively impact parental stress and mental health (Robinson & Neece, 2015). Increased parental stress, parental depressive symptoms, and other mental health concerns can exacerbate extant child behavior problems (Hauser et al., 2014). Parents of children with FXS tend to have higher rates of anxiety, stress, and depression than the general population (Abbeduto et al., 2004; Hartley, Seltzer, Head, & Abbeduto, 2012; Lewis et al., 2006), with between one fifth and nearly one half of mothers of children with FXS falling within the clinical range for depression (Baker, Seltzer, & Greenberg, 2012; Wheeler et al., 2007). Evidence also indicates that child problem behaviors negatively impact maternal mental health in biological mothers of children with FXS, and this can have an overall negative impact on the parent-child relationship (Fielding-Gebhardt, Warren, & Brady, 2019). Further, mothers of children with FXS appear to be more vulnerable to psychopathology compared to mothers of children with other disorders, such as Down syndrome (Lewis et al., 2006; Wheeler et al., 2007). This likely is partially due to their status as genetic carriers of the Fragile X permutation, but also the

result of increased pessimism about the future well-being of their child compared to mothers of children with other DDs (Lewis et al., 2006). It may be especially important that parent training for mothers of children with FXS incorporate CBT to combat this pessimistic thinking. PFI integrates PBS with “optimism training” (Seligman, 1998) in an effort to increase parents’ self-efficacy and optimism about their child’s ability to change. In a randomized clinical trial, parents of children with DD who displayed severely challenging behaviors reported significant improvements in their children’s problem behavior after receiving PFI compared to the group of parents who received PBS alone (Durand, Hieneman, Clarke, Wang, & Rinaldi, 2013). Although both the PFI and PBS groups showed reduced child problem behavior as rated by direct observation, the addition of CBT to the treatment resulted in a greater reduction in parent-reported child problem behavior. Families in the PFI group reported being better able to use the program’s strategies to manage child behavior problems than families in the PBS group alone, and they reported an increase in their child’s positive behavior rather than simply a reduction in their child’s problem behavior. This is an indication that, although both PBS and PFI improved negative behaviors, PFI was unique in improving parental perception of their child’s behavior. This indicates that PFI can be a particularly promising approach for mothers of children with FXS.

Parent Training for Children with Fragile X Syndrome

Children with FXS exhibit a wide range of often serious problem behaviors; however, the research on behavioral interventions (including behavioral parent training) is sparse. Behavioral intervention studies examining individuals with FXS – independent of other DDs – are even rarer. The research thus far has focused mostly on medical and

pharmacological interventions rather than behavioral or psychological interventions (Hall, 2009), perhaps because FXS is a genetic condition as opposed to a behaviorally defined disorder like autism.

Although a recent systematic review of the behavioral intervention literature in FXS (Moskowitz & Jones, 2015) identified a handful of studies in which parents were taught to implement behavioral interventions with their children, most of these studies included only one or two participants with FXS (e.g. O'Connor et al., 2003; O'Reilly et al., 2000; Schieltz et al., 2011; Wacker, Harding, & Berg, 2008) or included a heterogeneous group of individuals with DD in which the results for the participants with FXS were combined with the results of participants with other diagnoses (e.g. Feldman & Werner, 2002). In fact, in the review by Moskowitz and Jones, only three studies in which parents of children with FXS were trained to implement behavioral interventions to reduce problem behaviors (Kurtz, Chin, Robinson, O'Conner, & Hagopian, 2015; Moskowitz, Carr, & Durand, 2011) or sleep problems (Weiskop, Richdale, & Matthews, 2005) replicated intervention effects across at least three children with FXS.

Most recently, Vismara and colleagues (2018; 2019) piloted the initial feasibility and acceptability of parent-delivered Early Start Denver Model (P-ESDM), an intervention designed for children with ASD, with four mothers of children with FXS. They found that mothers improved in P-ESDM fidelity, implemented intervention goals, and rated the treatment as moderately to highly acceptable, although there were mixed results with regard to parent-reported improvement in children's socially appropriate and aberrant behaviors. In another recent study, Monlux and colleagues (2019) examined the effect of a function-based behavior analytic intervention delivered via the internet for

reducing problem behaviors in 10 boys with FXS, finding reductions of 78-95.3% in child problem behaviors and high parent integrity and acceptability ratings. Thus, the few studies that have been published show that behavioral parenting training interventions may hold promise for children with FXS.

Treatment Considerations

Parental cognitions about child behavior, including parental attributions, have been found to substantially influence the outcomes of parent training programs for child problem behaviors (Sawrikar & Dadds, 2017). Child-referent attributions refer to a parent's belief about how responsible their child is for their own behavior, while parent-referent attributions refer to a parent's belief about how responsible they as the parent are for their child's behavior (Mattek, Harris, & Fox, 2016). A recent study found that parents of children with ASD were more likely than parents of neurotypical children to believe their children could not control their problem behavior, which predicted their use of more lax parenting than the parents in the control group, which in turn, was associated with higher levels of child problem behavior for the children with ASD (Berliner, Moskowitz, Braconnier, & Chaplin, 2019). Targeting the beliefs of parents through cognitive restructuring may be particularly important for mothers of children with FXS, given that these parents often express the belief that their children's problem behavior is uncontrollable due to the role that biology plays in the syndrome. Indeed, more than 30% of parents reported that their child with FXS lacked the ability to control outbursts or displays of anxiety involving repetitive or self-injurious behavior (Woodcock, Oliver, & Humphreys, 2009). Although it is true that genetic variables influence the severity of expression in FXS (Hessl et al., 2008), environmental factors also impact problem

behavior displayed by children with FXS (Hall, DeBernardis, & Reiss, 2006; Langthorne et al., 2011; Machalicek et al., 2014). Parents' attributions that their child's problem behavior is uncontrollable and/or unchangeable might be a barrier to implementing behavioral interventions with fidelity, especially since behavioral interventions are predicated on the assumption that the environment impacts behavior.

One barrier to implementing behaviorally based parent-training programs for children with FXS is accessibility. The diagnostic rate of FXS is much lower than the 1 in 59 prevalence rate of ASD, the developmental disorder most commonly targeted by behavioral parent training (e.g., Christensen et al., 2016; Kasari, Gulsrud, Paparella, Helleman, & Berry, 2015; Warren et al., 2011). This means that many families of children with FXS live in locations where there are few providers who understand both the diagnosis and behavioral interventions. Families who have children with FXS report difficulty finding qualified service providers and transportation to and from appointments, and parents without access to qualified service providers are also more likely to rely on non-evidence-based treatments, despite having their child's best interests in mind (Elder et al., 2016). Studies have not specifically examined logistical barriers to treatment, but it is likely they are more pronounced for families of children with FXS given the lower prevalence of the diagnosis.

Some of these barriers can be addressed through the use of technology. Teletherapy is being explored as a potential alternative to in-person therapy across a range of diagnoses, from depression to ASD (Hickey, 2013). Several studies have found that parent training delivered via teletherapy was effective in teaching parents of children with ASD the skills (e.g. antecedent manipulation, functional communication training) to

manage child problem behaviors, and effective in decreasing problem behaviors (Benson et al., 2018; Machalicek, Lequia, & Raulston, 2016; Seuss et al., 2014; Vismara et al., 2018; Wacker et al., 2013b). Research also indicates that teletherapy-delivered interventions for ASD may be as effective as in-person therapy (Lindgren et al., 2016). Parent training studies that target reducing problem behaviors and/or increasing communication in children with FXS via teletherapy have only recently started to be published. Studies by McDuffie and colleagues (2016a, 2016b, 2018) found that a parent-mediated intervention for spoken language in boys with FXS, implemented through video-teleconferencing, substantially increased the boys' expressive language in the context of a joint activity. While these studies are promising, there is only one parent training study to date that has targeted problem behavior in individuals with FXS via telehealth: the aforementioned study by Monlux et al (2019). Although this study by Monlux and colleagues is an excellent starting point in terms of examining the effect of internet-delivered parent training on child problem behavior in FXS, Monlux et al. (2019) did not directly target mothers' mental health, an important variable that could affect child problem behavior, treatment dropout, intervention fidelity, intervention gains, and maintenance and generalization. Monlux and colleagues also limited their study to two behavioral interventions, functional communication training (FCT) and extinction, whereas the effects of training parents of children with FXS to implement a broader array of antecedent-based (prevention), replacement, and consequence-based (management) strategies via telehealth remain unknown.

Current Study

Although children with FXS commonly display a range of problem behaviors, including self-injury and aggression, and parents typically rate problem behavior as their greatest concern regarding their children with FXS (Hatton et al., 2002), there is little research examining the effectiveness of behavioral intervention – particularly behavioral parent training – to address problem behaviors in this population. Logistical issues (including access to service providers) and parents’ assumptions about the inability of their child to improve his or her behavior may detrimentally impact families seeking treatment or remaining in treatment. Given the effectiveness of PFI for treating problem behavior in children with DD (Durand et al., 2013) and the preliminary effectiveness of using telehealth to provide behavioral interventions for children with developmental disorders (including FXS), this study addressed three main research questions. First, this study evaluated whether PFI can reduce problem behavior in a child with FXS when PFI is delivered via teletherapy. Second, this study examined whether maternal perceptions of child behavior can change as a result of PFI. Finally, this study investigated whether parental mental health can improve after completing PFI, specifically whether maternal depressive symptoms and maternal stress improve from pre- to post-treatment.

Study

This study examined the potential effectiveness of PFI delivered via telehealth for reducing problem behaviors in a child with fragile X syndrome. There were three main hypotheses.

Hypothesis 1: PFI delivered via video conferencing would reduce child problem behavior (parent-reported problem behavior as well as directly observed problem behavior).

Hypothesis 2a: PFI would improve parental mental health by reducing parental stress as measured by the Parenting Stress Index, 4th Edition – Short Form (PSI-4-SF; Abidin, 2012) and the Questionnaire on Resource and Stress (Friedrich, 1983).

Hypothesis 2b: PFI would improve maternal mental health by reducing levels of parental depressive symptoms as measured by the Beck Depression Inventory, 2nd edition (BDI-II; Beck, Steer, & Brown, 1996) or the Center for Epidemiological Studies' Depression Scale, Revised (CESD-R; Radloff, 1977; Eaton et al., 2004).

Hypothesis 3: PFI would improve parental attributions by reducing inaccurate and/or child-referent attributions as measured by the Parental Attribution Questionnaire (PAQ; Walker, 1985).

Method

Participants

Participants were recruited through a variety of means, including a listserv for parents of children with FXS living in the New York City metropolitan area (the FXNY listserv), the national Fragile X parent listserv, and Facebook groups aimed at parents of children with FXS and related diagnoses. Recruitment was also conducted through the National Fragile X Foundation (NFXF) and in person at the International Fragile X Conference. To be included in the study, participating families could not currently be enrolled in parent training or have received Positive Behavior Support or a similar type of behavioral parent training intervention in the previous six months. Children on an unstable dose of medication were also excluded from participation.

The participant received an information packet via e-mail containing a letter describing the study, consent forms, and screening measures approved by the Institutional Review Board (IRB) at St. John's University. The packet was also available via traditional mail upon request. The screening materials included diagnoses, medications, and a brief history of prior parenting interventions or behavior therapy. It also included a pre-treatment assessments of problem behavior: the General Maladaptive Index (GMI) subsection of the Scales of Independent Behavior-Revised (SIB-R; Bruininks et al., 1996). Potential participants were also given measures of mental and emotional well-being including the Parenting Stress Index, 4th Edition – Short Form (PSF-4-SF; Abidin, 2012); the Questionnaire on Resources and Stress (QRS-F; Friedrich, 1983); the Beck Depression Inventory, 2nd Edition (BDI-II; Beck, Steer, & Brown, 1996); and the Center for Epidemiological Studies' Depression Scale, Revised (CESD-R; Radloff, 1977; Eaton

et al., 2004). Mothers were included in this study if they (a) had a son or daughter between the ages of three and 12 with a diagnosis of FXS (full mutation), residing in the home, who (b) exhibited problem behaviors as determined by a score of at least one standard deviation above the mean on the GMI index of the SIB-R (less than -21). Scores on the PSI-4-SF and the BDI-II were collected for analysis (pre-post treatment) but were not used in the selection process. A total of 40 potential participants expressed an interest in the study, 35 of whom either did not meet the inclusion criteria, did not return the screening materials, or declined to participate after learning more information about the study. Five interested parents met the inclusion criteria and were enrolled. Of the five enrolled mothers, three withdrew following difficulties in equipment and/or technology set up; one withdrew due to a reported reduction in her child's problem behavior prior to the beginning of the intervention. One participant, Julie (age 41 years) completed the treatment program for her son Peter (age 7 years), who was diagnosed with FXS and autism. Julie reported that Peter was minimally verbal, but was beginning to learn functional communication skills at the start of the study. Peter required assistance for the majority of his activities of daily living, including dressing, toileting, and using eating utensils.

Setting

Sessions were conducted via WebEx, a secure, web-based video conferencing client that allows for the recording of audio and video and meets HIPAA auditing requirements. This allowed the participant to complete her portion of the treatment from a wide range of locations, including her home or office. All PFI treatment sessions were conducted in the participant's home. The participant used her own computers and digital

cameras to participate in sessions from a location of her choosing. The investigator administered the modules from a private, locked room with the camera oriented on her face and a blank wall in the background. The sessions were delivered from a designated, password-protected research laptop. Recorded videos were stored in stored using Box®, a HIPAA-compliant software storage system, to be coded at a later date. Baseline videos were recorded using the participant's mobile phone, an iPhone 6. These videos were also uploaded Box® for later coding.

Experimental Design

To examine the effects of PFI on Peter's behavior and Julie's cognitions and emotions, a single-subject AB design was used (e.g., Sidman, 1960; Ledford & Gast, 2018). This type of design entails collecting baseline data on the dependent variables until the baseline data are relatively stable (A), then introducing the intervention (B). Baseline length varies depending on the length of time needed to achieve a stable baseline, in this case nine weeks. AB design is a commonly used approach in social science research and has been used in publications with small numbers of participants. For example, Monlux and colleagues (2019) conducted a study across ten participants with FXS using an AB design. Similarly, McManus and colleagues (2003) used an AB design to examine the effects of a toileting program for one six-year-old child with FXS using a two-week baseline (A), two-part 15-week intervention (B), and a follow-up assessment.

After the participant was screened and identified, the baseline phase (A) was started. Pre-treatment data for all dependent variables was collected for the participant at the outset of participation. Each week the participant collected baseline direct

observational data of her child's problem behavior. The participant remained in the baseline phase until the problem behavior data were stable, at which time the intervention phase (B) began. Direct observation problem behavior data was collected by the mother via video once per week. One additional in-depth measure of parent-rated problem behavior, as well as measures of maternal depression, stress, and attributions, were collected twice: once pre-treatment and once post-treatment. Two different measures were used to evaluate depression, stress, and problem behavior because of the limited research on families of children with FXS, differences in the designed use of available measures, and how often different measures are used in published research. Post-treatment follow-up began immediately following the final PFI session and lasted for a total of three sessions. The participant's first post-treatment observation occurred the first week following treatment. However, due to family emergencies, she was unable to record observational data during the next two weeks. Her second and third observations took place three and four weeks following the end of the treatment phase.

Measures

Scales of Independent Behavior, Revised (SIB-R)

Problem behaviors were also measured using the General Maladaptive Index (GMI) subsection of the SIB-R (Bruininks et al., 1996), a norm-reference measure of problem behavior. The SIB-R can be administered either via structured interview or as a self-report measure; in this instance, it was administered as self-report. For the purposes of this study, only the section measuring maladaptive behavior, the General Maladaptive Index (GMI), was used. GMI measures three areas of problem behavior: internalized maladaptive behavior; externalized maladaptive behavior; and asocial maladaptive

behavior. The rater is asked to rate both the frequency and severity/intensity of different behaviors in eight domains: behaviors that are hurtful to self; behaviors that are hurtful to others; behaviors that are destructive to property; disruptive behavior; unusual or repetitive habits; socially offensive behavior; withdrawal or inattention; and uncooperative behavior. Scores on this index place the child's behavior into one of five categories: very serious (–41 and below), serious (–40 to –31), moderately serious (–30 to –21), marginally serious (–20 to –11) and normal (–10 and above).

The SIB-R manual does not report internal consistency alphas for the GMI index alone. However, Bruininks and colleagues (1996) reported a test-retest reliability of .86 after two weeks, as well as acceptable inter-rater reliability and construct and criterion validity. Researchers have used the GMI with children who have developmental disabilities (Durand et al., 2013) and young adults with intellectual disabilities (McIntyre, Blacher, & Baker, 2002). The SIB-R GMI index was administered at pre-treatment screening and as a post-intervention follow-up measure.

Parenting Stress Index, 4th Edition, Short Form (PSI-4-SF)

Parental stress was evaluated using the PSI-4-SF (Abidin, 2012), which consists of 36 self-report items. It is used to measure stress level associated with parenting (Pérez-Padilla, Menéndez, & Lozano, 2015) and is designed to be used with children ranging from one month to 12 years of age. The PSI-4-SF is composed of three subscales: Parental Distress, Parent–Child Dysfunctional Interaction, and Difficult Child. Each domain is measured with 12 items, and each item is scored on a 5-point Likert scale ranging from 1 (strongly disagree) to 5 (strongly agree). The parent and child subscales combine to make the Total Stress scale. The PSI-4-SF is considered reliable, with both

the Parent and Child subscales showing test-retest reliability between .65 and .85 (Abidin, 2012) and is consistently reliable even at one-year follow-up (Haskett, Ahern, Ward, & Allaire, 2006). The PSI-4-SF also shows good internal consistency, with alphas ranging from .80 to .91. Haskett and colleagues (2006) report that the PSI-4-SF has good predictive validity for parent report of children's disruptive behaviors one year after the initial assessment. The PSI-4-SF was administered once as a pre-treatment and once a post-treatment follow-up measure.

Questionnaire on Resources and Stress (QRS-F)

Parental stress was also measured using the QRS-F (Friedrich, 1983), a 52-item measure evaluating parental stress designed for use with families of children who have disabilities. The QRS-SF was used in the original RCT on PFI with children with developmental disabilities by Durand et al (2013) and has also specifically been used in research on families of children with FXS (e.g., McCarthy, Cuskelly, van Kraayenoord & Cohen, 2006). Parents or caregivers respond True or False to questions about their expectations for their child, their own views of their family, and their attitude towards their child and family. Higher scores indicate a higher level of stress. The questions on the QRS-F can be factored into four categories: Parent and Family Problems, Pessimism, Child Characteristics, and Physical Incapacitation. For the purposes of this study, only the total score was used. The QRS-F has been found to have good internal consistency and reliability, with the reliability for the Total Score equaling .95 (Friedrich et al., 1983; Scott, Thompson & Sexton, 1989) and a mean score of 18.6 (SD=11.0; Friedrich et al., 1983). The QRS-F was administered once as both a pre-treatment measure and post-treatment follow-up.

Two measures were used to evaluate the construct of parental stress. The QRS-SF was used by Durand et al. in their 2013 paper on PFI. However, Durand and colleagues used it primarily to evaluate parental pessimism (which was not evaluated in this study) and noted some limitations in how the QRS-SF evaluates changes in parental views of their own self-efficacy. The PSI-4-SF has been used more widely in research on parental stress and was chosen as an additional measure in order to gather more robust data on parental stress.

Beck Depression Inventory, 2nd Edition (BDI-II)

The BDI-II (Beck, Steer, & Brown, 1996) is a 21-item measure of depressive symptoms, corresponding to DSM-IV criteria for depressive disorders (Smarr & Keefer, 2011). Each item presents four options, ranging in severity from 0 to 3, about a common depressive symptom. The items are then summed to produce a Total Score. The BDI-II has been designed for use with individuals ages 13 to 80 in both clinical and non-clinical samples (Beck, Steer, & Brown, 1996). A total score of 0-13 falls in the minimal range. A score between 14 and 19 indicates mild depressive symptoms; between 20 and 28 is moderate depressive symptoms; and between 29 and 63 is considered severe (Beck et al., 1996). The BDI-II is considered internally consistent, with a Chronbach's α between 0.92 and 0.93 (Smarr & Keefer, 2011). It has an acceptable test-retest reliability at one week, with a correlation of 0.93 between the two scores (Smarr & Keefer, 2011) and is positively correlated with other measures of depression (Beck et al., 1996). The BDI-II was given to the participating parent once as a pre-treatment measure and once as a post-treatment follow-up. It was used to evaluate maternal depression.

Center for Epidemiologic Studies Depression Scale (CES-D)

The CES-D (Radloff, 1977) is a 20-item measure of depression developed to be used in epidemiological studies of depression in the general population. The CES-D measures common depressive symptoms such as loss of appetite, anhedonia, and fatigue, occurring over the prior week. Because it was developed for use in the general population, it is short, free, and can be used by laypersons as well as mental health professionals. For these reasons, the CES-D has been widely used in research with individuals who have developmental disabilities, including people with FXS and their families (e.g., Lewis et al., 2006; Hunter et al., 2012). All 20 items on the CES-D are measured using a 3-point Likert-type scale, with zero indicating no symptomatology and a three indicating that the respondent experiences the symptom “most or all of the time.” Scores range from zero to 60, with higher scores indicating more symptomatology (Radloff, 1977). A score of 16 or more indicates the likely presence of clinically significant depression. Research has indicated that the CES-D has good reliability and validity and it is used both in research and clinical practice (Roberts, 1980; Zich, Atkisson, & Greenfield, 1990). Its internal consistency is reported to be high in both the general population ($\alpha = 0.85$) and a patient population ($\alpha = 0.90$; Radloff, 1977). Its test-retest reliability is reported to be moderate (0.45-0.70), which the creator of the measure indicates is part of its design as it is meant to measure symptoms over a relatively short period of time (Radloff, 1977). The CES-D was given to the participating parent once pre-treatment and once as a post-treatment measure.

Two measures were used to evaluate the construct of depression because although the BDI-II has a stronger psychometric background and is used in more published

research, the CES-D is designed for use in the general population rather than a clinical sample and has been used in research of people who are caregivers to children or adults with developmental disorders.

Parental Attribution Questionnaire (PAQ)

Parental attributions of child behavior were measured using the PAQ (Walker, 1985), a modified version of the Attribution and Control Questionnaire. The PAQ has previously been used in research on children with ASD and other developmental disorders (e.g., Whittingham et al., 2008). For the purposes of this study, only the PAQ's "bad behavior" scenario was used; parents are asked to consider a time in which their child exhibited problem behavior, then answer 12 follow-up questions using a 5-point Likert scale ranging from "Strongly Disagree" to "Strongly Agree." Responses fall under child-referent versus parent-referent attributions, and higher scores reflect more internal, stable, and controllable causes (Hartley et al., 2013). Child-referent attributions refer to a parent's belief about how responsible their child is for his/her own problem behavior. Parent-referent attributions refer to a parent's belief in his/her own causal role in the child's problem behavior. The PAQ has good internal consistency (0.79-0.89; Hartley, Schaidle, & Burnson, 2013). The participating mother was given the PAQ once pre-treatment and once as a post-treatment follow-up measure.

Observed Problem Behavior

In addition to subjective parent-reported problem behavior, problem behaviors were also assessed objectively using direct observation from video recordings. The participating mother recorded her child using a mobile phone during a naturalistic routine or situation that usually results in problem behaviors – in this case, a trip to a

supermarket. The videos ranged in duration from 5 minutes and 59 seconds to 12 minutes and 56 seconds, but each video encompassed the entirety of the targeted routine. This video data was collected and uploaded in the same week during baseline, intervention, and post-treatment follow-up. No feedback was given to the parent regarding the videos themselves; rather, the participating parent discussed the problem situation during the intervention (as directed by the PFI manual). The recorded videos were scored by the principal investigator using 10-second partial-interval time sampling. By collecting ongoing observational data throughout treatment, the intervention sessions that correlated with a significant reduction in observed problem behaviors could be recognized.

Social Validity Measure

Following the completion of PFI, the principal investigator administered a 29-item self-report “Parent Satisfaction Questionnaire” in order to evaluate the intervention’s social validity. This survey used a 5-point Likert rating with options ranging from 1 (strongly disagree) to 5 (strongly agree). Questions 6-9 in Part A were reverse-scored to help control for response bias. Part A asked the participant about the social validity of the specific routine targeted in PFI. Part B asked about treatment acceptability, in particular regarding the process of receiving the intervention via telehealth (e.g., the use of the web camera, WebEx, and privacy or security concerns). Part C asked the participant about social validity with regard to her life in general, outside of the specific targeted routine (e.g., “I am more optimistic about my child’s future,” “In general, my child’s behavior plan fits my family’s needs, priorities, and situation.”) This measure was administered in part due to the limited research on parenting interventions for FXS, in particular web-

based parenting interventions for FXS; qualitative data addressing perceived positive and negative aspects of treatment were collected to help guide future research on the topic.

Materials

The present study followed the treatment manual *Helping Parents with Challenging Children: Positive Family Intervention Facilitator Guide* (Durand & Hieneman, 2008). A number of materials were required as part of the PFI program. These forms are included in the PFI parent workbook (Durand & Hieneman, 2008a). The participant was given an electronic copy of the relevant workbook pages prior to each session. The workbook is 135 pages and pairs with the PFI facilitator's guide (Durand & Hieneman, 2008b). Additional copies of the forms in the workbook were available upon request.

The participant also needed to have access to a video-capable mobile phone and a laptop or desktop computer capable of supporting WebEx software.

Procedure

Prior to beginning data collection, the participating mother was administered the screening measures (SIB-R, BDI-II, CESD-R, QRS-F, PSI-4-SF, PAQ). After these measures were returned and scored, the participant underwent a pre-treatment evaluation of problem behavior. This evaluation began with a semi-structured phone interview conducted by the principle investigator (PI) about common situations that were challenging for her child. This discussion resulted in the PI and the participating mother choosing one target problematic situation to record for baseline, intervention, and post-intervention follow-up data. The situation needed to be (1) routine, occurring at least once per week; (2) recordable via mobile phone (e.g., not a situation in which recording

would be impossible or unethical, such as the classroom or while the child was bathing); (3) predictably challenging to complete due to problem behaviors. The participant was coached on how to collect this data by the PI. Baseline recording lasted for nine sessions, ranging from six minutes 50 seconds to 11 minutes and 20 seconds in duration.

Following the completion of baseline data collection, the participant was instructed on the installation of WebEx software on her phone and home computer. She received the intervention via web camera on her laptop and cell phone. The participant provided her own internet connection. The investigator delivered the intervention via a designated research laptop in a locked room using a web camera and WebEx. The sessions were recorded and stored on the WebEx website until they were transferred to the research laptop.

PFI consists of eight individually-administered once-per-week sessions, each lasting approximately 90 minutes. Only the mother participated in the 8 sessions; children or other caregivers were not present. Each module was presented to the parent using WebEx software. Workbook pages were emailed to the parent immediately prior to each relevant web session, or mailed to the participant up to a week in advance. The PI, a clinical psychology doctoral candidate with a Masters of Art in Clinical Psychology and a background in CBT and ABA, conducted all sessions.

PFI focuses on teaching both the principles of PBS and CBT. PFI delivers the modules in a pre-determined order. Session One (Introduction and Goal-Setting) focuses on setting goals, identifying problematic situations, and identifying parental cognitions. Session Two (Gathering Information on Challenging Behavior) concentrates on teaching the parent to collect data to conduct a functional behavior assessment (FBA), which

involves identifying the antecedents and consequences of problem behaviors, along with identifying the consequences of parental beliefs (parents' "self-talk"). Session Three (Analyzing Data and Plan Design) teaches parents to analyze the FBA data to identify patterns, then brainstorming possible intervention strategies based on the FBA data, along with cognitive restructuring of parents' negative self-talk. Session Four (Using Prevention Strategies) focuses on selecting prevention strategies to address specific child problem behaviors, using distraction to interrupt parents' pessimistic or unhelpful self-talk, and cognitive restructuring of negative parental thoughts. Session Five (Using Consequences) covers selecting consequence-based intervention strategies to manage the child's problem behaviors and using affirmations to target and replace negative thoughts. Session Six (Replacing Challenging Behavior With Appropriate Alternatives) works on replacing problem behavior with appropriate alternative behaviors along with practicing skills to identify and change parental negative self-talk. Session Seven (Implementing the Strategies) focuses on the implementation of the selected prevention, replacement, and management strategies for problem behaviors and continues work on parents managing their self-talk. Finally, Session Eight (Monitoring the Results) focuses on results: how to track and monitor them and how to maintain the treatment gains. Each session includes homework assignments to allow for the practice of strategies at home, as well as case examples and practice applying these tools. Post-treatment, the participant continued to submit weekly videos of the same problem situation identified in pre-treatment for a minimum of three sessions. After completing PFI, the PI administered a measure of social validity.

Treatment Integrity and Interobserver Agreement

The author's doctoral dissertation mentor reviewed a portion of each video-recorded PFI session, and feedback on these sessions was provided in weekly supervision. One hundred percent of the 90-minute modules were coded for procedural fidelity from video by both the PI and two trained research assistants to confirm that all components of each given module were addressed in each session and to ensure fidelity to the model on the part of the PI. The second raters were a trained graduate student and a trained undergraduate student who used a procedural fidelity assessment created by Durand et al., (2013), which consisted of a checklist of 10 to 13 items to be covered in each session. Intervention fidelity was calculated across all of the nine sessions and was found to be 100% (number of items completed divided by total number of listed items, multiplied by 100).

Interobserver agreement (IOA) was calculated using an interval-by-interval method in which the percentage of intervals where both observers agree was analyzed using an agreement/total calculation (see Davis, Durand, Fuentes, Dacus, & Blenden, 2014) for 33% of baseline, intervention, and post-treatment sessions. Sessions included in IOA were coded by either a graduate or an undergraduate assistant. The primary observer (C.M.) and secondary observer (an undergraduate-level research assistant) independently coded 33% of all baseline sessions. Another secondary observer (a graduate-level research assistant) independently double-coded 33% of all treatment and post-treatment sessions. Both research assistants were trained using the same procedures using practice videos until an acceptable level of reliability (greater than 90%) was achieved before coding began using video-recordings of the participant's problem behavior. IOA for

problem behaviors during baseline, treatment, and intervention sessions was 87.79% (number of agreements divided by number of agreements plus disagreements, multiplied by 100)

Results

Direct observations of problem behavior were analyzed using visual analysis as the primary analysis and mean baseline reduction (MBLR) and Tau-U (Parker, Vannest, Davis & Sauber, 2011) as secondary analyses. In an AB single-subject design, Tau-U examines the non-overlap of data points between the baseline phase (A) and the intervention phase (B) and combines it with the trend of the intervention (B) phase. Tau-U also allows for one to control for baseline (A) trend. A Tau-U of .65 or lower is considered a minimal or small effect; scores between .66 and .92 are categorized as a medium to high effect, and .93 to 1.0 can be interpreted as a large or strong effect (Parker et al., 2011; Rakap, 2015; Rispoli et al., 2013).

The PFI program begins with three sessions dedicated to laying the groundwork for behavior change; parents do not implement any behavioral strategies until Session Four. To balance this, mean-baseline reduction (MBLR) was also calculated by subtracting the average of the last three treatment data points from the last three baseline data points, dividing the result by the mean of the last three baseline points, and multiplying by 100 (Olive & Smith, 2005). Since MBLR as a nonparametric technique seemed most appropriate for our study, we decided to supplement our analysis with Tau-U to provide some estimate of effect size. MBLR does not provide cutoff scores, and was used in conjunction with Tau-U to counterbalance the potential of Tau-U being skewed by outliers.

Maternal depression, maternal stress, child- and parent-referent attributions, and parent-reported measures of problem behavior were analyzed using the percentage of change from pre- to post-treatment for each corresponding measure and participant (see Table 1).

Parent-Reported Child Problem Behavior

Parent-reported child problem behavior was measured in two ways: the Scales of Independent Behavior—Revised, General Maladaptive Index (SIB-R) and direct observation via video recording. Julie’s pre-treatment ratings of her child’s problem behavior according to the SIB-R fell into the Serious range (GMI= -31). Her post-treatment SIB-R score fell into the Marginally Serious range (GMI= -17), a reduction of 48.48% on this measure.

Direct Observation of Child Problem Behavior

Observational data from the baseline, intervention, and post-treatment phases were collected and the frequency of problem behaviors was assessed. Over the course of 20 weeks, 19 observational videos were collected. Julie was unable to perform the grocery store routine the 5th week of the treatment phase (week 15 of the study) due to both an illness afflicting her son and the Christmas and New Years holidays. The mean frequency of observed problem behavior for the grocery store routine during baseline was 51.21% ($SD=22.08$, range=4–72.90). The mean frequency of observed problem behavior for the last three data points in the intervention phase was 1.33% ($SD=0.94$, range=0–2) (see Figure 1), with a 97.29% mean baseline reduction in the frequency of problem behavior from baseline to intervention.

A visual inspection of the observational data was also conducted. Single-case design data can be visually evaluated in terms of level, immediacy, variability, and trend (Riley-Tillman & Burns, 2011). When evaluating level, the level of the baseline phase is compared to the level of the treatment phase. Because the primary goal of intervention research is to change a target behavior following a treatment, the level between the baseline and treatment phases should show a noticeable change during a visual inspection. Mean differences can also be compared during an evaluation of level. Figure 1 displays the results of Peter's observational data. Peter's baseline data show a high level of variability, indicating that the duration and frequency of his problem behaviors during the shopping routine was irregular, with an overall average frequency of 51.21% ($SD=22.08$) for all baseline sessions. During the visual analysis, the level of Peter's problem behavior during the shopping routine clearly dropped during the treatment phase to an average frequency of 30.85% ($SD=38.30$) for all eight treatment sessions. The effect of PFI is not immediate; there is no clear drop in observed problem behavior until Session Four of PFI (in week four), which corresponds to the PFI module in which parents begin to actively implement strategies for reducing problem behavior. The variability of Peter's data during the treatment phase is high when all data points are considered together; however, if one considers only the data points following Week 4, the variability is much lower ($M=2.25$; $SD=2.06$). Trend is less clear; it appears as though Peter's data during the treatment phase is trending downwards. However, his data during the post-treatment follow up phase is more variable and does not have a clear trend, although the level is lower than baseline.

The data were also evaluated using a Tau-U with corrected baseline procedure (Parker et al., 2011; Parker, Vannest, & Davis, 2014). Tau-U ranges from 0 to 1. Because of the clear visual drop in level of observed problem behaviors beginning in Session Four of the treatment, Tau-U was calculated twice: once for phase A and B in their entirety, and once for phase A and the data points from Session Four onward in phase B. Tau-U was calculated using an online calculator designed by Parker and colleagues (see <http://www.singlecaseresearch.org/calculators/tau-u>).

Before calculating Tau-U, baseline trend was evaluated to determine whether it needed to be considered in the Tau-U calculations. The results for baseline trend were calculated using an online Baseline Corrected Tau calculator (Tarlow, 2016). The results indicated that it was reasonable to assume a stable baseline ($\text{Tau} = -0.357, p = 0.266$). Tau-U was calculated and results indicated that the overall improvement trend for the complete phase A and phase B equaled .32 (90% CI [-0.82; 0.18]; $p = 0.30$), which indicates a weak trend that does not reach statistical significance. Improvement trend for phase A compared to phase B from Session Four onward yielded a larger result of .93 (90% CI [-1;-0.33]; $p < .05$), indicating that, when visual analysis was taken into consideration (as well as the content of the sessions), a large effect size was reached.

Parental Stress

This study hypothesized that parenting stress, measured by the PSI-4-SF, would decrease following the intervention. Julie's ratings of parental stress reduced from a total score of 105 to 83, a decrease of 20.95% (see Table 1). This indicates that Julie's self-report of her own parenting stress reduced from the Clinically Significant range before treatment to the High Stress range following treatment.

It was further hypothesized that participants' stress would decrease following the intervention as measured by the Questionnaire on Resources and Stress (QRS-F). The QRS-F has a mean score of approximately 18.6 (SD=11.0; Friedrich et al., 1983). Julie's initial score on the QRS-F was 31, over one standard deviation above average. She obtained a post-intervention score of 28, a reduction of 9.68%. This reduction in parental stress is notably less than the reduction in stress as measured by the PSI-4-SF, and still falls approximately one standard deviation above the mean.

Parental Depression

Julie obtained an initial score of 12 on the BDI-II, which falls into the Minimal Depression category. She obtained a follow-up score of 2, still placing her in the Minimal Depression category, though with an 83.33% reduction. Julie's scores on the CES-D were also reduced, although not as markedly. Julie obtained a pre-treatment depression score of 31 as measured by the CES-D, and a post-treatment depression score of 16; a 48.39% reduction in depressive symptoms as evaluated by this measure. Scores of less than 16 are considered not clinically significant for depression; higher scores indicate more symptomatology. Julie's ratings on the CES-D indicate an overall reduction in depressive symptoms, but still fall in the clinically significant range.

Parental Attributions

Child-Referent Attributions

This study hypothesized that the participant would report a decrease in internal, stable, and controllable child-referent attributions as measured by the Parental Attribution Questionnaire (PAQ) following the intervention. The participant was given the PAQ pre- and post-treatment. In the domain of child-referent attributions, Julie reported a 33.33%

decrease in attributions to internal causes; a 33.33% decrease in attributions to stable causes; and a 37.5% decrease in attributions to controllable causes (see Table 1). This indicates that, post-treatment, Julie attributed her child's problem behaviors to less internal, less stable, and less controllable causes when compared to her attributions pre-treatment.

Parent-Referent Attributions

It was further hypothesized that participants would report a decrease in parent-causal attributions as measured by the PAQ. In the domain of parent-referent attributions, Julie reported a slight reduction (33.33%) in attributions to internal causes; a medium reduction (66.67%) in attributions to stable causes; and a no change in attributions to controllable causes (0.00%) (see Table 1). This indicates that, post-treatment, Julie was less likely to attribute her child's problem behavior to causes internal and stable to her as a parent, and equally likely to attribute her child's behavior to things that she, as a parent, could control.

Social Validity of PFI delivered via WebEx

Table 2 displays Julie's ratings on the Parent Satisfaction Questionnaire completed following the end of the study. Part A of the Parent Satisfaction Questionnaire asked questions related to the completion of PFI and the implementation of the chosen target routine. Julie indicated that, overall, she found the intervention moderately helpful for the target routine (grocery shopping) and that the improvement seen during this specific situation had a large impact on her sense of control as a parent. Her average rating of overall improvement in the targeted routine was 4.07 out of five ($SD = 0.28$).

Part B asked questions pertaining to the treatment acceptability of PFI, including the use of WebEx software and quality of treatment. Julie indicated “Strongly Agree” to all questions in this section, including those pertaining to the voice and video quality of the calls, the knowledge of the clinician, and the comfort and ease of receiving treatment in this manner. These responses indicate that Julie was very satisfied with the method of treatment delivery itself. Her average rating of treatment acceptability was 5 out of five ($SD = 0$).

Part C asked the respondent to think about their life in general rather than just the specific target routine that was chosen. Julie responded in more varied ways to the questions in Part C than to the questions in Part B. Her responses indicate that she felt more able to establish goals, gained more understanding of her child’s behavior, and felt more confident and optimistic about her ability to work with her child and her child’s future in general. Her results also indicated that she did not see much improvement in her child’s overall replacement skills and was unsure about the effect this intervention would have on her overall quality of life. Her average rating of overall improvement was 4.08 out of five ($SD = 0.95$).

Discussion

This study investigated the effect of Positive Family Intervention (Durand et al., 2013), delivered via teletherapy, on the problem behaviors of a child with FXS and the mental health of his mother. As hypothesized, Peter exhibited a moderate improvement in directly observed problem behavior according to the results of the visual analysis and Tau-U. Visual inspection of the data shows that the largest drop in observed problem behavior occurred after Session Four, the first module in which parents begin implementing the tools for behavioral change. The decrease is visually clear and distinct. Unfortunately, the post-treatment observational data, which began immediately following the end of the treatment phase, contained three observations, and lasted a total of four weeks, is less decisive in that the data are more variable. However, it is important to note that, at the same time as the follow-up data was being collected, Julie's father-in-law was hospitalized for a serious medical issue. Julie's father-in-law's illness began in the second week post-treatment, which resulted in a delay of data collection by one week and may have impacted her child's mood and behavior as well as Julie's own mood, thoughts, and behaviors, and her overall level of stress. These factors could have contributed to the high degree of variability between the three post-treatment observations, particularly between the first and third observation compared to observation two. Julie's ratings of her child's problem behavior according to the SIB-R GMI index also showed a decrease from pre- to post-treatment of 48.48% from Serious to Marginally Serious. This score still indicates the presence of some problem behavior post-treatment, although it represented a substantial reduction from pre-treatment, and Julie noted at the end of treatment that some self-injurious behavior had begun to emerge in situations outside of the grocery

store routine. The SIB-R GMI index is a more general/broad measure of problem behavior than the direct observation data from videos, which only examined problem behavior in one target situation (i.e., grocery shopping). As such, it makes sense that Julie's child's problem behavior reduced more markedly in one of the specific situations that was directly targeted by PFI than it did in general/overall in areas not directly targeted by the treatment, although the improvement from Serious to Marginally Serious suggests that some generalization may have occurred from the target situation to other areas of life.

It is interesting to note the pattern of decrease in problem behaviors as measured by direct observation. Peter's instances of problem behavior during the baseline phase were highly variable, but on average occurred at over 50% of recorded intervals. His treatment-phase data show a large drop-off following Session Four which, as mentioned previously, is the session in which parents begin to implement strategies identified in the first three sessions. This decrease is clear, stable, and marked. It is worth asking whether the first three PFI sessions set the stage for this noticeable decrease, what particular components may have been most essential, and how the results might look were the aspects of treatment to be teased apart.

At the beginning of the study, it was hypothesized that the mother's depressive symptoms would be reduced at the end of PFI, theoretically as a function of the CBT component of the program. Symptoms of depression as measured by both the BDI-II and the CES-D decreased for Julie. However, Julie's scores on the BDI-II decreased more than her scores on the CES-D. These measures are scaled differently and have typically been used with different populations: the BDI-II is often used in mental health facilities

as a depression screener, whereas the CES-D was developed for use with the general population. This difference in applied population could explain the variation in intensity of depressive symptoms between the two measures. Julie's initial rating on the BDI-II fell into the Minimal Symptoms of Depression category; her decrease in score, while large, still falls in the same group. Julie's initial score on the CES-D was a 31, which indicates a much more severe presence of depressive symptoms. This could be due to the populations for which these measures were designed; the BDI-II was designed for use with a clinical population, while the CES-D is normed for use with the general population. Julie obtained a post-treatment score of 16 on the CES-D, the lowest possible score one can receive while still technically meeting the cut-off for likely depression. This indicates that her depressive symptoms were likely reduced, but not eliminated. Although this reduction did not meet significance according to the CES-D cut-offs, it is likely to indicate some level of clinical significance.

This study further hypothesized that ratings of parental stress regarding child problem behavior would decrease as a result of PFI. The CBT component of PFI helps parents to identify negative/pessimistic, inaccurate, and/or unhelpful thoughts and work to change those thoughts to more optimistic or helpful cognitions. This appears to have been the case for Julie. Julie's ratings of parental stress as measured by the PFI-4-SF and the QRS-F decreased as well but, again, the magnitude of the decrease differed substantially between measures. Julie's score on the PSI-4-SF decreased by 21%, bringing her from the Clinically Significant range to the High Stress range, while her ratings on the QRS-F lowered by 10%. The QRS-F is a continuous measure of stress and thus does not have distinct categories or cutoffs; however, the reported above-average

score is 18.6 with a standard deviation of 11. Julie's score on the QRS-SF did not decrease by even one standard deviation, and was still elevated post-treatment compared to the mean. However, as Durand et al (2013) noted, the QRS-SF was designed to assess parents' overall perceptions rather than measure changes in parents' attitudes about their child's ability to change specific behaviors in targeted situations, which could explain why Julie's QRS-SF scores showed limited change from pre- to post-treatment. Further, while the PSI-4-SF has been used in studies on families of children who have a developmental disability, it was not specifically designed for use with that population. The QRS-SF, on the other hand, was designed for use with families of people who have a developmental disability. Thus, it is possible that the QRS-F is detecting stress specifically associated with having a child who has a disability, a type of parenting pressure that could be more challenging to reduce compared to the general stress of having children. Additionally, the PSI-4-SF uses a 5-point Likert scale, while the QRS-F only allows True or False response options. This could affect the sensitivity of the QRS-SF, leading it to show less apparent change over the course of treatment.

Julie's dysfunctional parental attributions as measured by the PAQ reduced for both child-referent and parent-referent beliefs. Previous research indicates that parents' dysfunctional attributions are positively associated with depressive symptoms and child problem behavior, among other characteristics (e.g., Snarr et al., 2009; Wilson, Gardner, Burton & Leung, 2006). Following treatment, Julie was less likely than before treatment to believe that Peter's problem behavior was caused by factors that were internal and stable to her child. Post-treatment, she was also less likely to believe that Peter could control his behavior compared to her pre-treatment beliefs. This latter finding about

control may not necessarily be a positive/desirable outcome, given that recent research indicates that parents believing that their children with ASD have either too much control (e.g., “He’s doing this on purpose”) or too little control over their challenging behavior (e.g., “He can’t help it because he has autism”) could lead to less effective methods of discipline (Berliner, Moskowitz, Braconnier, & Chaplin, 2019). In terms of behavioral intervention, it is important that parents believe their child have *some* amount of control over their behavior in order to believe that they are capable of changing it. Following treatment, Julie was less likely to believe her child’s problem behavior was caused by factors that were internal and stable to her (the parent), though just as likely to believe that she as the parent could control the behavior, the latter of which may be a realistic belief and may even be a helpful belief as long as it does not lead to self-blame. It was hypothesized that the CBT component of PFI would reduce dysfunctional attributions, and this appears to be the case for Julie.

Limitations and Future Directions

There are a number of limitations to this study. First, given that the present study used an AB design, which is considered a pre-experimental or quasi-experimental type of single-subject design, we cannot definitively establish a functional relationship between PFI and the outcomes due to threats to internal validity. Therefore, any changes we observed from pre- to post-treatment cannot be conclusively attributed to PFI as opposed to just the passage of time, history, maturation, etc. That said, research shows that problem behaviors such as aggression and self-injury are highly likely to persist over time in individuals with FXS (e.g., Crawford, Karakatsani, Singla, & Oliver, 2019), so it

is unlikely that the child's problem behavior would have simply improved on its own without intervention.

Second, in addition to these threats to internal validity, the number of participants was limited to only one mother of a child with FXS and, therefore, the results of this study cannot be generalized to other mothers of children with the same diagnosis, nor to all children with FXS and problem behaviors (which limits our external validity). Given that the aim of this pilot study was to provide initial support for the feasibility, acceptability, and effectiveness of delivering PFI in a telehealth format for mothers of children with FXS, an important step for future research is to examine PFI using a more rigorous experimental design with a larger group of parents. Larger samples are needed in order to draw conclusions regarding the effectiveness of internet-delivered PFI for children with FXS. Another shortcoming of this particular study is that only mothers were recruited. While on the one hand this can be viewed as a limitation that fathers were not included, on the other hand the decision was intentional, given that fathers are not carriers for FXS, and so do not experience the same feelings of guilt, self-blame, and depression that mothers of children with FXS and other X-linked conditions often experience (e.g., James et al., 2006; Kay & Kingston, 2002; McConkie-Rosell et al., 1997, 2000, 2001). However, fathers and other caregivers are also responsible for parenting their children with FXS, and little research has investigated the use of behavioral interventions with non-maternal caregivers in this population. This is a gap in the research that should be addressed.

It also must be noted that this study contained several potentially confounding variables. With all the different aspects of treatment, it is impossible to determine which

components affected the outcomes. Relatedly, another important limitation of this study involves the pitfalls of conducting research remotely. Obtaining materials from the participant, including observational videos and screening packets, proved challenging throughout the research. Additionally, collecting observation videos was reportedly challenging for parents, as there were issues both with obtaining the videos themselves and uploading the videos for coding. The former problem resulted in the loss of at least two potential participants. That said, there are many advantages to web-based services – convenience, cost, and access being primary.

Another important limitation is that we did not examine generalization and maintenance. In particular, the lack of long-term follow up is an issue that future research should address. As mentioned previously, Julie's child started to engage in self-injurious behavior around the time the PFI intervention ended. This was a new problem behavior that had not been specifically targeted by the intervention. Although Julie had been taught skills to identify the function of new problem behaviors, it is unknown whether Julie was able to adapt or generalize the skills learned during PFI to this new situation. Follow-up at six, 12, or 24 weeks post-treatment would have given a clearer picture of the lasting impact of PFI on behaviors that were not specifically targeted during the study itself. Given the lack of research into the effect of behavioral interventions on children with FXS, these types of data could prove highly valuable. Additionally, the data collected suggest that, for this participant, parental cognitions, stress, and depressive symptoms appeared to change following the completion of PFI, although we cannot rule out other variables that may have contributed to that change with just an AB design. In spite of the positive changes that Julie reported and that were seen in the direct observation data, we

do not know whether those changes were long-lasting, whether depressive symptoms would have continued to improve over time, or how the change in cognitions would apply (or not apply) to new situations. It is certainly possible that, with the passage of time, we might see a re-emergence of Julie's dysfunctional attributions, unhelpful or inaccurate cognitions, and depressive symptoms. In addition, while the therapist's procedural fidelity was evaluated by an observer, the fidelity of the participant was not assessed. Given the challenges inherent in implementing behavioral strategies, future research ought to examine parents' fidelity to the intervention plan developed in PFI. Another limitation involves the measures themselves. While some of the measures were designed for use with parents or caregivers of children with disabilities (e.g., the QRS-F), none have been specifically evaluated for use with families of children who have FXS. This may account for some of the variation in pre- to post-treatment changes between instruments supposedly measuring the same construct. Similarly, the measure of parental attribution used in this study (PAQ, Bad Behavior Scenario) does not specifically ask about disability-specific attributions. It may be assessing broad attributions made by parents generally (e.g., "my child cannot control his behavior") but not those specific to having a child with a genetic syndrome such as FXS (e.g., "my child cannot control his behavior because he has FXS"). The PAQ is also a fairly recent measure and is comprised of only two questions per measured construct. Extant measures simply do not have the specificity required to understand the finer grain of parental attributions, stress, or depression as it relates to the evaluation of parents with children who have developmental disabilities, and FXS in particular. There may be better, more targeted ways of addressing thoughts related to FXS-specific attributions.

The methods of data collection are another limitation of this study. Due to the number of measures and because of the challenges involved in collecting data remotely, the majority of the data were collected only twice: once at pre-treatment and once at post-treatment. This makes most robust statistical analyses impossible. While it would have been preferable to collect data at each session, the burden on participants was already thought to be quite high, and it is not clear whether these measures would be sensitive to change from week to week. Therefore, it was decided that most of the measures (BDI-II, CES-D, PFI-4-SF, QRS-F, SIB-R, PAQ) would only be collected at two time points rather than at each session.

This study also had several significant strengths. In the only other existing behavioral parent training study delivered via teletherapy with parents of children with FXS (Monlux et al., 2019), rates of problem behavior were only recorded during telehealth treatment sessions rather than in typical daily-life routines/situations outside of the sessions. One major strength of the present study is that direct observation of problem behavior was collected in naturalistic routines/settings outside of the PFI session during one of the pre-determined challenging situations that typically occurred in daily life. Further, the intervention plan was implemented by a natural intervention agent (i.e., the child's mother) in a naturalistic setting (i.e., the grocery store). Thus, the present study demonstrates high ecological validity, meaning that the intervention can be successfully used in real-world situations. The combination of parent-report via the SIB-R and the direct observation of problem behavior provide a level of converging support about the trajectory of Peter's problem behavior that other studies with more participants typically lack. Additional strengths include the use of a manualized intervention program designed

to target both problem behavior and parental cognitions that may be contributing to the maintenance of problem behavior. This is important given that, to date, no published behavioral intervention studies targeting problem behavior in children with FXS have directly addressed both the child's problem behavior and the parents' thoughts and feelings.

Although the results of the present study are encouraging, additional research is needed examining the use of PFI with caregivers of individuals with FXS. Researchers should examine PFI for families of children with FXS under more controlled conditions. As discussed, the sheer number of variables in this study makes it challenging to draw conclusions about what specific aspects were beneficial to treatment. Looking at the components of PFI under more controlled circumstances with a sharper focus on the different aspects of the treatment could provide valuable knowledge about which aspects contribute to positive change and which may not.

Given that the present study examined PFI that was delivered remotely, in-person PFI is another avenue that still remains to be explored with parents of children with FXS. Although another study found that ABA for children with ASD was equally effective in reducing problem behaviors if it was delivered in-person, through clinic-based teletherapy, or through home-based teletherapy (Lindgren et al., 2016), it is possible that in-person PFI might be even more effective than PFI delivered via teletherapy for parents of youth with FXS, or at least for some parents. Determining which parents of children with FXS (or ASD or other developmental disabilities, for that matter) may need the in-person support versus the parents for whom telehealth PFI will be sufficient is a critical direction for future research. In addition to in-person versus remote treatment, other

parent training programs and behavioral intervention strategies should also be explored with individuals with FXS, given the limited research on behavioral interventions – particularly behavioral parent training – designed to address problem behavior in this population. Future research should examine whether PFI (which both directly targets the child’s problem behavior using PBS as well as the parent’s thoughts and feelings through CBT) is more effective at reducing problem behavior than PBS-alone (or some other type of behavioral parent training on its own without CBT) for parents of children with FXS.

One issue that became clear during the course of this research was the challenges potentially participating parents faced in setting up and accurately using the required technology. Many of the challenges, for example the reported difficulties in uploading videos to Box.com, are specific to the research context. There are several alterations that could be made when transitioning PFI to a clinical setting. First, as mentioned above eliminating the use of Box.com or other video hosting platform to evaluate problem behavior. PFI traditionally does not include direct observation of problem behavior, and this component was added for research purposes. Collecting and uploading these videos was reportedly challenging. Second, reducing the number of measures clients need to complete from six to perhaps one or two would ease the burden placed on families enrolled in treatment. Finally, assisting clients in the set-up and use of the technological components of this intervention would alleviate many of the reported challenges in the use of technology itself.

Conclusion

In general, the present study adds to the limited body of literature that examines the use of behavioral parenting interventions on improving challenging behavior in those

with FXS. It also provides promising data regarding the impact of combining aspects of CBT into PBS when addressing the problem behaviors of a child with FXS, including decreasing parental pessimism, unhelpful beliefs, and dysfunctional attributions, and improving rational beliefs and perceptions of the severity and frequency of their child's challenging behavior. These benefits extend beyond those already demonstrated by Durand et al. (2013). Teaching cognitive strategies may help improve parents' ability to implement behavioral interventions with fidelity, cope with problematic situations, and generate more optimistic views about their children and their future. It also provided some preliminary information regarding the effectiveness of using telehealth to deliver behavioral parent training interventions to parents of children with FXS. Further research is needed; however incorporating these findings into clinical practice when working with families of children with FXS could help make parenting interventions more effective by addressing parental mood, attributions, and beliefs while also tackling challenging behavior.

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Table 1.

Means and Percentage of Change for Parental Stress, Attributions, Depression, and Ratings of Problem Behavior

Measure	Normative Mean	Pre-Treatment	Post-Treatment	% Change	Descriptor
PSI-4-SF (Stress)		105 (Clinically Significant)	83 (High Stress)	-20.95%	Improvement
QRS-F (Stress)	M = 18.6 SD = 11.0	31	28	-9.68%	Slight Improvement
PAQ Parent-Referent (Attributions)					
Internal	M = 7.25 SD = .20	6	4	-33.33%	Less Internal
Stable	M = 7.00 SD = .23	6	4	-33.33%	Less Stable
Controllable	M = 5.17 SD = .24	8	5	-37.50%	Less Controllable
PAQ Child-Referent (Attributions)					
Internal	M = 4.39 SD = .26	6	4	-33.33%	Less Internal
Stable	M = 6.92 SD = .21	6	2	-66.67%	Less Stable
Controllable	M = 5.23 SD = .24	6	6	0.00%	No Change
BDI-II (Depression)		12 (Minimal Symptoms)	2 (Minimal Symptoms)	-83.33%	Slight Improvement
CES-D (Depression)		31 (Clinically Significant)	16 (Clinically Significant)	-48.39	Slight Improvement
SIB-R GMI (Behavior)		-33 (Serious)	-17 (Marginally Serious)	+48.48%	Improvement

Table 2.

Items and Raw Scores Social Validity Measure

Item	Rating
<i>Part A</i>	
1. I was able to establish goals for this routine that will benefit my child and family.	4
2. I have a greater understanding of what affects my child's behavior in this routine.	4
3a. I have developed a clear plan for my child's behavior in this routine, with strategies to prevent problems	4
3b. I have developed a clear plan for my child's behavior in this routine, with strategies to encourage positive behavior	4
3c. I have developed a clear plan for my child's behavior in this routine, with strategies to respond more effectively	4
4. My child's behavior plan for this routine fits my family's needs, priorities, and situation.	4
5. I am able to implement the strategies in my child's plan consistently in this routine.	4
6. My child's problem behavior has decreased during this routine in comparison to before PFI began.	4
7. My child's positive behavior (e.g., replacement skill) has increased during this routine in comparison to before PFI began.	4
8. This routine is more enjoyable now in comparison to before PFI began.	4
9. This routine is easier now in comparison to before PFI began.	4
10. I use the strategies that I have learned for this routine in other routines or situations.	4
11. Improvement in this routine has made me feel more in control as a parent.	5
<i>Part B</i>	
1. Receiving this program via webcam was comfortable.	5
2. The voice quality of the sessions was acceptable.	5
3. The video quality of the sessions was acceptable.	5
4. Participating in this intervention using telehealth was convenient.	5
5. I received the same quality of treatment via telehealth as I would have with an in-person provider.	5
6. My privacy was well protected.	5
7. My clinician was knowledgeable about the intervention.	5

Table 2.

(continued)

Item	Rating
<i>Part C</i>	
1. As a result of my involvement in the PFI project I was able to establish goals (in general) that will benefit my child and family.	5
2. I have a greater understanding of what affects my child's behavior (<u>in general</u>).	5
3a. I have developed a clear plan for my child's behavior in general (in a variety of situations), with strategies to prevent problems.	4
3b. I have developed a clear plan for my child's behavior in general (in a variety of situations), with strategies to encourage positive behavior.	4
3c. I have developed a clear plan for my child's behavior in general (in a variety of situations), with strategies to respond more effectively.	4
4. In general, my child's behavior plan fits my family's needs, priorities, and situation.	5
5. In general, I am able to implement prevention, replacement, and management strategies consistently.	4
6. Since participating in Positive Family Intervention my child's problem behavior has decreased in general (in situations other than the target routine).	4
7. Since participating in Positive Family Intervention my child's positive behavior (e.g., replacement skills) has increased in general (in situations other than the target routine). In other words, my child has learned skills in the target routine that he now uses in other situations.	2
8. My child's quality of life (e.g., community participation, quality of relationships, general satisfaction) has improved in general. In other words, we can go more places, do more things, and are generally happier.	3
9. My family's quality of life has improved in general (e.g., see above).	3
10. I feel more confident in my ability to work with my child in general.	5
11. I am more optimistic about my child and his or her future in general.	5

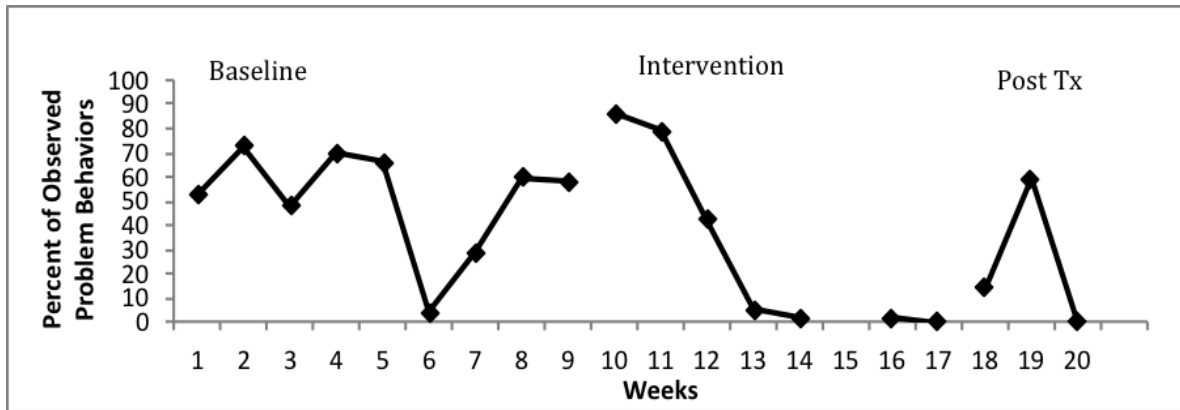


Figure 1. *Percentage of observed operationally-defined problem behavior during pre-treatment, treatment, and post-treatment for Participant 1 (Julie). Note: No data for week 15.*

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