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ORIGINAL PAPER

Extending the Parent-Delivered Early Start Denver Model to Young Children with Fragile X Syndrome

Laurie A. Vismara[1] · Carolyn E. B. McCormick[2] · Rebecca Shields[1,3] · David Hessl[1,3]

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Abstract

This is the first study to evaluate an autism intervention model, the parent-delivered Early Start Denver Model (P-ESDM), for young children with fragile X syndrome (FXS), a known genetic disorder associated with autism spectrum disorder. Four parent–child dyads participated in a low-intensity, parent coaching model of the P-ESDM to evaluate initial efficacy and acceptability. Parents improved in P-ESDM fidelity, implemented intervention goals to increase child learning, and found the experience moderately to highly acceptable. Visual examination and Baseline Corrected Tau effect sizes revealed mixed results across child measures. Findings suggest a potential therapeutic opportunity in need of larger, well-controlled studies of P-ESDM and other interventions for families of young children with FXS who face limited empirically-supported intervention options.

Keywords Fragile X syndrome · Autism spectrum disorder · Parent-mediated · Early intervention · Early Start Denver Model

Fragile X syndrome (FXS) is the main hereditary cause of intellectual disability (ID) and the most common monogenic cause of autism spectrum disorders (ASD) (Bailey et al. 2008; Farzin and Koldewyn 2014; Hagerman et al. 2008; Hernandez et al. 2009; Lozano et al. 2016; Wheeler et al. 2015). Children with a dual diagnosis exhibit poorer developmental outcomes (Hatton et al. 2006; Roberts et al. 2007), including late-onset language milestones, weaker communication and social skills, lower adaptive behavior scores, greater behavior problems, and more significant cognitive impairment than boys with FXS only or those with idiopathic ASD (Bailey et al. 2000, 2001; Kau et al. 2004; Hernandez et al. 2009; Rogers et al. 2001, 2003a, b). Likewise, there is considerable burden to families (Chevreul et al. 2015). A study examining patients with FXS in the USA reported that approximately one-third of caregivers had seen a professional for anxiety, stress, or depression during the previous year, and one-quarter took medication to address these symptoms (Bailey et al. 2012). Caregiver burden was highly associated with problem behavior, and almost one-third of the caregivers had been injured by their child with FXS at least once in the past year.

Despite pressing need, intervention research on children FXS and ASD is scarce (Hagerman et al. 2009; Hall 2009; Reiss and Hall 2007) often with an N of 1 for each behavioral strategy tested or with school-aged and adolescent males making it difficult to draw conclusions about its effectiveness for toddler-aged children (McDuffie et al. 2016a, b, 2018; Moskowitz and Jones 2015). The American Academy of Pediatrics and Committee on Genetics provides clinical guidelines on genetic testing and recommended examinations for well-care visits but no prescribed course of action for treatment and how to support affected families (Hersh

✉ Laurie A. Vismara
laurie.vismara@gmail.com

Carolyn E. B. McCormick
mccorm37@purdue.edu

Rebecca Shields
rhshields@ucdavis.edu

David Hessl
drhessl@ucdavis.edu

1 MIND Institute, University of California Davis Medical Center, 2825 50th Street, Sacramento, CA 95817, USA

2 Human Development and Family Studies, Purdue University, Fowler Memorial House, Rm. 202 1200 W. State Street, West Lafayette, IN 47907-2055, USA

3 Department of Psychiatry and Behavioral Sciences, University of California Davis Medical Center, 2230 Stockton Blvd., Sacramento, CA 95817, USA

and Saul 2011; Moeschler and Shevell 2014). The lack of empirically validated behavioral intervention models present a major problem for interventionists and educators working with young children with FXS or with FXS and ASD. Consequently, parents may rely on intervention methods with little to no research supporting efficacy for their children.

What we know about early intervention and brain development in ASD research suggests that the way in which young children interact with their environment affects brain connections and neural responses (Drew et al. 2002; Harris and Handleman 2000). Early intervention plays a crucial role in this process in shaping the child's developing brain to be receptive to the social information presented in order to accelerate language learning and learning rates in young children with a variety of developmental disorders (Wallace and Rogers 2010). The most prominent comprehensive early intervention models in use and under investigation for young children with ASD adopt a naturalistic approach fused with behavioral and developmentally-sensitive teaching practices called Naturalistic, Developmental, Behavioral Interventions (NDBIs; Bradshaw et al. 2015; Schreibman et al. 2015). NDBIs rely on this transactional model of development (Sameroff and Chandler 1975) wherein the quality and responsiveness of environmental factors influence child developmental gains and is highly mediated by how parents provide for, interact with, and talk to their children. Several RCTs have examined the effectiveness of NDBIs implemented by parents given children's young ages at the start of treatment and the advantage of immediate access it provides for parents to embed the intervention inside activities they can do, themselves. Parents are able to meet their goals to help their children engage, communicate, and learn as part of their typical caretaking and interactive routines (Baranek et al. 2015; Carter et al. 2011; Green et al. 2015; Kasari et al. 2010; Schertz et al. 2013; Wetherby et al. 2014).

The parent-delivered Early Start Denver Model (P-ESDM; Rogers et al. 2012a) is an emerging NDBI that integrates applied behavior analysis (ABA) principles with a developmental, relationship-based approach for sensitive, responsive teaching focused on children's interests, emotional regulation capacity, and a developmental perspective of how skills and behaviors unfold for infant to preschool-aged children with ASD (Rogers and Dawson 2010; Rogers et al. 2014). The first RCT of a 12-week P-ESDM program for families of toddler-aged children with ASD showed no advantage over a "treatment-as-usual" community intervention group with respect to parent implementation or its effect on child outcomes (Rogers et al. 2012b). Subsequent coaching enhancements made to dosage, multimodal learning tools, and motivational interviewing techniques in the next RCT revealed greater gains in parent interaction skills with this enhanced version compared to the initial P-ESDM approach, as well as a significant relationship between parent change in interaction and rate of child improvement on developmental and autism symptoms (Rogers et al. 2018). Promising parent–child gains are also suggestive from single subject and controlled designs of coaching parents in the P-ESDM via video-conferencing sessions in families' home and access to online self-guided learning materials (Vismara et al. 2013, 2016). These studies suggest possible treatment gains from P-ESDM coaching for families of young children with ASD. To our knowledge, parent-mediated NDBI models, such as the P-ESDM, have not been studied as a viable treatment for FXS with or without ASD.

The present study attempted to pilot the initial feasibility and acceptability of coaching parents to use the enhanced version of P-ESDM with their child. Since the scope of the study was preliminary and exploratory, we used a single-subject design to address the following questions with four parent–child dyads affected by FXS with or without ASD:

1. Can parents use the P-ESDM with their child as measured by their fidelity of implementation and frequency to which they successfully taught intervention goals to their child?
2. Do parent gains in the P-ESDM improve children's use of spontaneous communication, joint attention initiations, and associated symptoms of FXS (e.g., anxiety, social withdrawal, arousal needs)?
3. Do parent–child gains persist across the intervention, maintenance, and post-assessment phase?
4. Do parents appear to enjoy using the P-ESDM with their child per their report and naïve ratings during sessions?

Methods

Participants

Four parent–child dyads participated in the study. The families either were referred to the study as part of their ongoing participation with the center's clinical and/or research services or contacted the first author after reading the study description posted on the center's website. To participate in the study, children had to meet the following criteria: (a) diagnosed with the *FMR1* full mutation of FXS; (b) between the ages of 18–48 months; (c) ambulatory; (d) a developmental quotient of 35 or above on the Mullen Scales of Early Learning (MSEL; Mullen 1995) and; (d) at least one primary caretaker to attend all sessions. FXS was confirmed using *FMR1* DNA testing at the Center or by review of prior DNA test results prior to study enrollment. All children were evaluated for ASD at the start of the study by the last author. Other community-based intervention services were not restricted but monitored for participating families, particularly because of the low-intensity nature of the current study.

The first four parent–child dyads that met the inclusion criteria were selected for enrollment. The study was approved by the Institutional Review Board (IRB) and adhered to the Health Insurance Portability and Accountability Act (HIPAA) in response to privacy, security, and electronic transaction guidelines. No adverse effects of the study occurred. Once consented and enrolled, parents completed a general demographic questionnaire concerning family composition, size and disabilities, annual gross incomes, educational levels and occupations, and the type and amount of other educational, behavioral, and/or medical services provided to children outside of this study. Parents could skip any question or item they felt uncomfortable answering.

Table 1 outlines key parent–child characteristics. Three of the four children were males with the full mutation of FXS and confirmed ASD diagnoses whereas the other child was female with the full mutation of FXS alone. The three male children had no or limited verbal speech and some communicative sign language (i.e., no more than 10 signs) whereas the female child communicated in short phrased speech to indicate needs and wants. Mothers were identified as the primary caregivers to participate in the study, although fathers attended occasional sessions.

Experimental Design and Procedure

A single-subject, multiple baseline design across four parent–child dyads (Hersen and Barlow 1976) examined parent implementation of the P-ESDM intervention strategies and its effect on child social-communicative behaviors. Parent–child dyads participated in 2 days of baseline assessments, 12 weekly 1.5 h intervention sessions, six bimonthly 1.5 h maintenance sessions, and finally one 2-h post-assessment approximately 2 weeks from the last maintenance session to reevaluate parent–child outcomes.

Baseline

Baseline occurred across two consecutive two-hour days at the center in order to administer standardized assessments, parent-report measures, and parent–child play activities. Parent–child dyads were randomly assigned to different predetermined baseline probes (Edgington 1996) to evaluate skills in a timely manner and not delay the start of critical intervention. In these probes, parents were instructed to play with their child, as they typically would do at home with any of the toys they or their child selected. The testing room and toys were not used during intervention or maintenance sessions.

P-ESDM Intervention

Parent–child dyads 1 and 2 participated in intervention and maintenance sessions at the center whereas the other two families participated remotely via video-conferencing due to out-of-state residence for dyad 3 and the first author's relocation during the study for dyad 4. Video-conferencing sessions occurred through the Citrix program GoToMeeting® in accordance with IRB approval and HIPAA regulations to protect the confidentiality and integrity of families' health information through a combination of encryption, strong access control, and other protection methods. Parents connected from their home, usually in their family room or child's bedroom, with a laptop, tablet, or computer and web-camera. Time was set aside before the first intervention session to practice and ensure that

Table 1 Parent–child demographics ($n = 4$)

	Dyad 1	Dyad 2	Dyad 3	Dyad 4
Child's chronological age in months	27	40	25	28
Child's gender	Male	Female	Male	Male
Child's ethnicity	African-American	Caucasian	Hispanic	Caucasian
Child's diagnosis	FXS & ASD	FXS w/o ASD	FXS & ASD	FXS & ASD
Child's medication per day	None	None	3–5 mg melatonin, 3.75 Zyrtec	2.5 mg sertraline
Additional services in hours per week	1 ST, 60 OT	10 inclusive PS 7 special education PS	1 ST, 0.5 OT, 1 2 ST	0.5 PT, 1 DV, 1 FT
Mother's chronological age in years	32	32	43	44
Mother's education	College	High school	College	Graduate
Mother's employment status	Full-time	None	Full-time	Part-time
Mother's marital status	Married	Married	Married	Married
Number of siblings	1	1	1	1

FXS Fragile X syndrome, *ASD* autism spectrum disorder, *w/o* without, *ST* speech and language therapy, *OT* occupational therapy, *PS* preschool, *PT* physical therapy, *DV* developmental visit from an early childhood specialist, *FT* feeding therapy audio–video equipment and internet connections worked for the therapist and families to see and hear one another.

The P-ESDM (Rogers et al. 2012a) is the parent-delivered intervention of the ESDM and follows the same science of applied behavior analysis and developmental, relationship-based intervention of the ESDM (Rogers and Dawson 2010). Its content and approach to working with families develop moments of learning inside the daily interactions and activities that make up a young child's life so as to teach and strengthen different areas of development. These everyday moments, whether picking out what to wear for the day, looking at a picture book, playing peek-a-boo, or splashing water in the kitchen sink, provide opportunities for the parent to emphasize the social function of language and to help beginning "talkers" understand nonverbal communication and imitation as foundations for verbal language. The P-ESDM sessions intended to teach parents how to use topics from, *An Early Start for Your Child with Autism* (Rogers et al. 2012a), to target multiple skills across different areas of development within any particular activity. These topics are attention and motivation, sensory social routines, joint activity routines, nonverbal communication, imitation, joint attention, speech development, functional and symbolic play skills, and the teaching techniques and learning contingencies of applied behavior analysis. Parents received copies of the book in the first intervention session.

The first author delivered twelve weekly1.5-h sessions to each of the four parent–child dyads. Sessions followed the same format and conformed fully to the detailed parent coaching manual (Rogers et al. 2012a), curriculum, parent fidelity of implementation measure (Rogers and Dawson 2010), and a defined fidelity of implementation measure for the coach (Rogers and Vismara 2013) regardless of in-person versus video-conferencing coaching. In the first session, the coach and parent jointly identified between 10 child learning goals (see Table 2) from initial assessments on which progress data were gathered during each session. Sessions 2–10 started with a brief check-in with parents about intervention usage and experiences since the previous session followed by activities of the parent or child's choice differing from those used in baseline probes before coaching proceeded. Activities were coded as individual probes to evaluate parent–child responses to the P-ESDM intervention. Coaching then focused on typical family interaction routines (books, feeding, dressing or changing, water activities, toy play, social play) for parents to practice an intervention topic and strategies related to parent–child goals and behaviors observed in the first activity followed with reflection and planning of how to carry over to home life and with other caretakers. Weeks 11–12 involved review of taught skills and setting parent–child goals to revisit in bimonthly maintenance sessions.

Maintenance

Parent–child dyads were seen twice-per-month (either at the center or via video-conferencing) for 1.5-h sessions each for 3 months, totaling six sessions. Maintenance sessions followed the same coaching structure as intervention sessions. After greeting the family, the coach asked the parent to engage their child in one or more activities that represented something they had been practicing since their last session (e.g., introducing a new game or skill). No coaching occurred during the activities. Activities were coded as individual probes to evaluate parent–child outcomes measures to the P-ESDM intervention. Following the parent–child activity, the remainder of the session focused on whichever P-ESDM topic(s) or particular learning goals parents selected or seemed important to strengthen based on earlier observations from the coach followed by practice, reflection, and planning next steps for parents to continue their practice and goals at home.

Post-assessment

Following the last maintenance session, parent–child dyads returned to the center for one final 2-h post-assessment. The last author re-administered standardized testing (except for the ADOS-T), parent-report measures, and parent–child interactive probes. This visit took place in a different room than where intervention occurred for those families whose sessions occurred at the center. Parents were given the same set of toys used during baseline and instructed again to play with their children as they would normally do so.

Therapist Training

The first author served as the therapist delivering the P-ESDM intervention and maintenance sessions. The first author co-developed and tested the intervention in-person and via video-conferencing in previously published studies. The first author's coaching skills were monitored on a bi-monthly basis through random selection of one videotaped session per family by another qualified therapist trained by the model's developers. The first author demonstrated 85% or higher fidelity on all coaching domains, including a set of practice and privacy guidelines when using online forums to deliver intervention (see Trepal et al. 2007, for more information) as assessed by Likert-based scores (1–4); otherwise supervision by a trained colleague was available until fidelity reached a level of competent practice (i.e., "3" or higher) over two consecutive sessions.

Table 2 Children's intervention goals

Child 1	Child 2
Responds with a related gesture or vocalization to parent language or play action	Responds on topic to parent questions
Completes two-step parent-given direction	Replies with the correct verb tense to describe an action
Spontaneously vocalizes at least 4 different consonants	Asks for help without protest
Combines two communicative behaviors to request continuation of activity	Replies with "I don't know" in appropriate context
Imitates parent-modeled play action or gesture	Asks, "What's that?" in appropriate context
Completes 3 or more functional play actions with toys	Labels opposites across 3 or more categories (large/small, tall/short)
Completes at least one pretend play action with a prop (hairbrush, sunglasses, phone)	Pretends to use an object as something else during make-believe play on self and parent
Uses communicative action (raises arm to be picked up, takes parent hand, points to go elsewhere) with parent to cope with unfamiliar or excitable situation	Follows board-game rules
Waits or comes back to parent when asked	Informs parent of potty needs before accident
Accepts or chooses a toy or activity to do without protest when 1st choice not possible	Follows bedtime routine without protest
Child 3	**Child 4**
Completes new play action modeled or offered by parent	Responds with a related gesture or vocalization to parent language or play action
Completes two-step parent-given instruction	Looks at parent when name is called to see what parent has
Spontaneously vocalizes at least 5 different consonants to request, refuse, or share	Completes two-step parent-given direction
At least 10 verbalizations contain a consonant-vowel-consonant-vowel sequence	Spontaneously names the item or action in request to parent
Looks at parent and gestures to show toy or item when asked	Spontaneously vocalizes with 3 or more words per utterance to parent
Imitates parent-modeled play action or gesture	Imitates parent-modeled play action or gesture
Accepts or chooses a toy or activity to do without protest when 1st choice not possible	Uses communicative gestures, vocalizations, and/or play-related actions to request that activity continue
Remains calm with a quiet voice, keeps body to himself and clothes on when frustrated	Looks, smiles, and vocalizes with 3 or more word utterances to share enjoyment with parent
Participates in at least 2 parent-chosen activities for at least 5–10 min	Pretends to use an object as something else during make-believe play
Shares toys or materials with peers and sibling without protest	Accepts or chooses a toy or activity to do without protest or self-injurious behavior when 1st choice not possible

Measures

Coding Procedures

Videos were coded in random order across baseline, intervention, maintenance, and post-assessment sessions to minimize expectations of parent–child progress. Four raters naïve to the intervention practiced coding on session tapes not involved with this study until they achieved 85% or higher agreement across at least 3 different tapes with an expert coder. Raters then worked in pairs to code the defined measures below. Reliability was assessed across 10% of videos across pairs with the expert coder. Raters adhered to defined coding definitions and rating systems research tested and published thus far with this intervention model (Dawson et al. 2010; Estes et al. 2014; Rogers et al. 2012b, 2018; Vismara et al. 2013, 2016; Vivanti and Dissanayake 2016; Vivanti et al. 2013). The second author scored standardized tests.

Parent Fidelity

Parent fidelity was rated by multiple probes per session across the four conditions using the published and previously research-tested ESDM Fidelity Rating System (Rogers and Dawson 2010), a Likert-based, 5-point rating system of 13 adult behaviors. Scores range from one (i.e., no competence) to five (i.e., high competence) with a total mean score of 80% or scores of 4 or greater indicating skilled and greater use of the intervention practices. Inter-rater agreement using weighted kappas was $\kappa = 0.55$ across 18 cases and $\kappa = 0.34$ across 36 cases.

Parent-Implemented Goals

The frequency with which parents successfully taught specific skills from their child's list of intervention goals (see Table 2) was examined across intervention and maintenance sessions. Raters examined the extent to which parents created an event, action, or circumstance (i.e., the antecedent) to target a specific child response or skill from the list of intervention goals (e.g., parent holds out a toy for the child to spontaneously request or names and points to a picture in a book for the child to see) followed by whether or not the child successfully responded to the learning opportunity (e.g., child spontaneously names the toy that the parent is holding or follows the parent's point to look at the picture). Codes resulted in a percentage of targeted intervention goals defined by the number of successful opportunities divided by number of missed opportunities. Inter-rater reliability involved raters identifying the same parental action as targeting a specific learning objective and whether its outcome (i.e., for the child to demonstrate the behavior defined in the goal) was met. The mean agreement between raters was 92.86%, with a range of 68.42–100.00%.

Parent Session Engagement

A measure was developed by the first author to assess parent engagement behaviors specific to the P-ESDM coaching and session structure (i.e., checking in, warm-up activity, topic introduction, coaching activities, reflection/evaluation, goal planning and generalization) during intervention, maintenance, and post-assessment sessions. Raters used a 1–5 (low versus high) global scale to assess the extent to which parents: (a) engaged in dialogue with the coach (e.g., asked and answered questions, shared progress, initiated information); (b) set goals and parameters for measuring their progress toward desired outcomes; (c) actively participated in the intervention during sessions (e.g., initiated activity ideas, asked for additional practice, selected goals to target); and (d) appeared comfortable and at ease with the coach (e.g., smiling, relaxed body positioning). Agreement was defined as raters globally selecting the same rating for each item after watching the full session. Interrater reliability between two coders as assessed with an ICC was 0.56.

Parent Self-Rating of Intervention Acceptance

The first author developed six statements rated 1–5 (low versus high) for parents to rate their perception of confidence, interest, and enjoyment using the P-ESDM during intervention and maintenance sessions. Statements were: (a) "I know how to use the P-ESDM topic within the daily activities and routines that happen with my child at home"; (b) "I know how to use the P-ESDM topic in settings or places other than my home"; (c) "I enjoy using the P-ESDM topic with my child"; (d) "My child responds positively (e.g., participates in the activity, does not leave the activity, smiles) to the P-ESDM topic"; (e) "I am able to help my child learn new behaviors and skills with the P-ESDM topic"; and (f) "I feel confident using this P-ESDM topic".

Coded Child Behaviors

We applied behavioral coding schemas used in previous research (omitted for blind review) to assess children's response to parents' use of intervention across conditions. Probes were transcribed and scored only for child initiation of: (a) functional verbal utterances of single words or approximations (echolalic or unintelligible utterances were excluded) directed toward the parent with body orientation to request or comment about an item or action; and (b) nonverbal joint attention behaviors of eye gaze alternation with or without gestures (i.e., giving, showing, pointing) directed to the parent to share interest or enjoyment in the activity. Only spontaneous verbal communication and joint attention initiations were used in analyses to reflect independent and not prompted child behaviors. Interrater reliability assessed with ICCs ranged from 0.96 to 0.98 for joint attention and 0.74–0.97 for spontaneous words.

Ratings of Socially Appropriate and Aberrant Behaviors

Global ratings of child behaviors from a 1–5 Likert-based scale (1 = rare; 3 = occasional; 5 = frequent) evaluated the extent of socially appropriate communicative behaviors and challenging, aberrant behaviors that the child directed to the parent across all conditions. Socially appropriate behaviors involved the child directing eye contact, vocalizations, gestural communication, engagement, and interest to the parent. Aberrant behaviors included continual staring or visual fixation to objects or items in the room, repetitive or stereotypic motor mannerisms, heightened awareness, uneasiness, or other signs of anxiety. Raters selecting the same score per behavior constituted an agreement. Interrater reliability was assessed with ICCs of 0.68 for socially appropriate behaviors and 0.52 for aberrant behaviors.

Standardized Child Assessments

Standardized assessments were administered by the last author, a clinical psychologist who did not participate with the P-ESDM intervention or maintenance sessions. The Autism Diagnostic Observation Schedule for Toddlers (ADOS-T; Lord et al. 2012) and Autism Diagnostic Observation Schedule, second edition (ADOS-2; Lord et al. 2002) are semi structured direct observational measures to evaluate social-communication symptoms in young children suspected of having ASD. The ADOS was administered based on child chronological age and expressive language abilities only at baseline to determine the presence and severity of ASD symptoms, whereas the other listed measures occurred during both baseline and post-assessment. The ADOS was excluded from the post-assessment battery because of several high-quality, high intensity ASD early intervention studies showing that it was not a sensitive measure of treatment related lessening of ASD symptoms (e.g., Dawson et al. 2010, 2012; Rogers et al. 2012b). Thus, we did not expect great change from this measure given our study involved a low-intensity parent mediated intervention.

The Mullen Scales of Early Learning (MSEL; Mullen 1995) assesses the cognitive functioning of young children from birth to 68 months and is based on the child's responses to activities prepared by the examiner. It measures five skill domains, including receptive and expressive language, fine and gross motor, and visual reception. The measure yields a composite score reflecting overall cognitive ability with a mean of 100 and standard deviation of 15.

The Vineland Adaptive Behavior Scales, Second Edition (VABS II; Sparrow et al. 2005) is a measure of adaptive behavior from birth to adulthood. Parents reported their perceptions of child functioning across communication, socialization, daily living, and motor skills.

MacArthur-Bates Communicative Development Inventory: Words and Gestures (MCDI; Fenson et al. 2007) is a 396-word parent report vocabulary checklist used to capture the expressive words, gestures, and receptive vocabulary a child has demonstrated in the past week.

Results

Data Analysis

Effect of intervention on parent and child outcomes was first analyzed by visually reviewing graphical displays of the outcome variables for changes in variability, range, and trend. All outcome variables were then analyzed using Baseline Corrected Tau, a non-parametric rank correlation effect size (Tarlow 2017). Compared to other methods of calculating effect sizes in single-subject research, Baseline Corrected Tau has values bounded between − 1 and + 1, and a decision tree is used to only correct for baseline trends when there is sufficient evidence to do so (i.e. a statistically significant monotonic baseline trend). Effect sizes were calculated using an online calculator (ktarlow.com/stats/tau/; Tarlow 2016). We report individual effect sizes, standard error, p-values, and whether we corrected for a baseline trend. Of note some parent outcome variables (i.e., parent-implemented goals, parent self-rating of intervention acceptance, and parent session engagement) were compared between intervention and maintenance to post-assessment sessions. Because we expected a positive increase in these variables across intervention, we did not correct for trends if present.

Parent Fidelity

Figure 1a displays parents' skill level using the P-ESDM intervention with children across the four intervention conditions. Across baseline probes, Parent 1's scores remained below the fidelity criteria of 4.00. During intervention, Parent 1's fidelity appeared to improve with scores fluctuating slightly above and below 4.00. Across maintenance and post-assessment, scores were mostly above 4.00 (i.e., 9/12 probes) indicating that Parent 1 implemented the intervention with integrity across the majority of probes. During nine baseline probes, Parent 2 had variable scores with all but one well below 4.00. Parent 2's skills demonstrated less variability during intervention but the majority of scores were still below 4.00. Of 11 maintenance and post-assessment probes, scores were very close to the fidelity criterion but only two were at or above 4.00, indicating an inconsistent fidelity performance for Parent 2. Parent 3 showed some increase in skills across baseline but performance did not maintain at fidelity. Skills across intervention probes also continued to be mostly below fidelity criterion. By maintenance and post-assessment, Parent 3's skills achieved fidelity particularly in latter sessions, indicating more consistent fidelity implementation. Lastly, Parent 4 consistently demonstrated below fidelity skills during baseline. Across intervention, Parent 4 skills increased gradually with probes above 4.00 towards the end of the phase. Across 11 maintenance and post-assessment probes, Parent 4 met fidelity with the exception of one probe. Effect sizes reported in Table 3 confirmed visual analyses of the data. Parents overall demonstrated significant improvement in fidelity from baseline to maintenance and post-assessment.

Parent-Implemented Goals

Figure 1b represents the percentage of successfully targeted intervention goals by parents across intervention and maintenance sessions. Parent 1 did not begin intervention effectively implementing intervention goals (mean = 0.07, SD = 0.10, range 0.00–0.29). Starting at intervention session 8, Parent 1 began a steady increase in the proportion of implemented goals that was continued across maintenance sessions (mean = 0.67, SD = 0.13, range 0.44–0.83). Parent 2 began the intervention implementing a low proportion of goals and began to steadily increase the proportion of implemented goals at the sixth intervention session (mean = 0.19. SD = 0.17, range 0.00–0.43) that was continued across maintenance sessions (mean = 0.61,

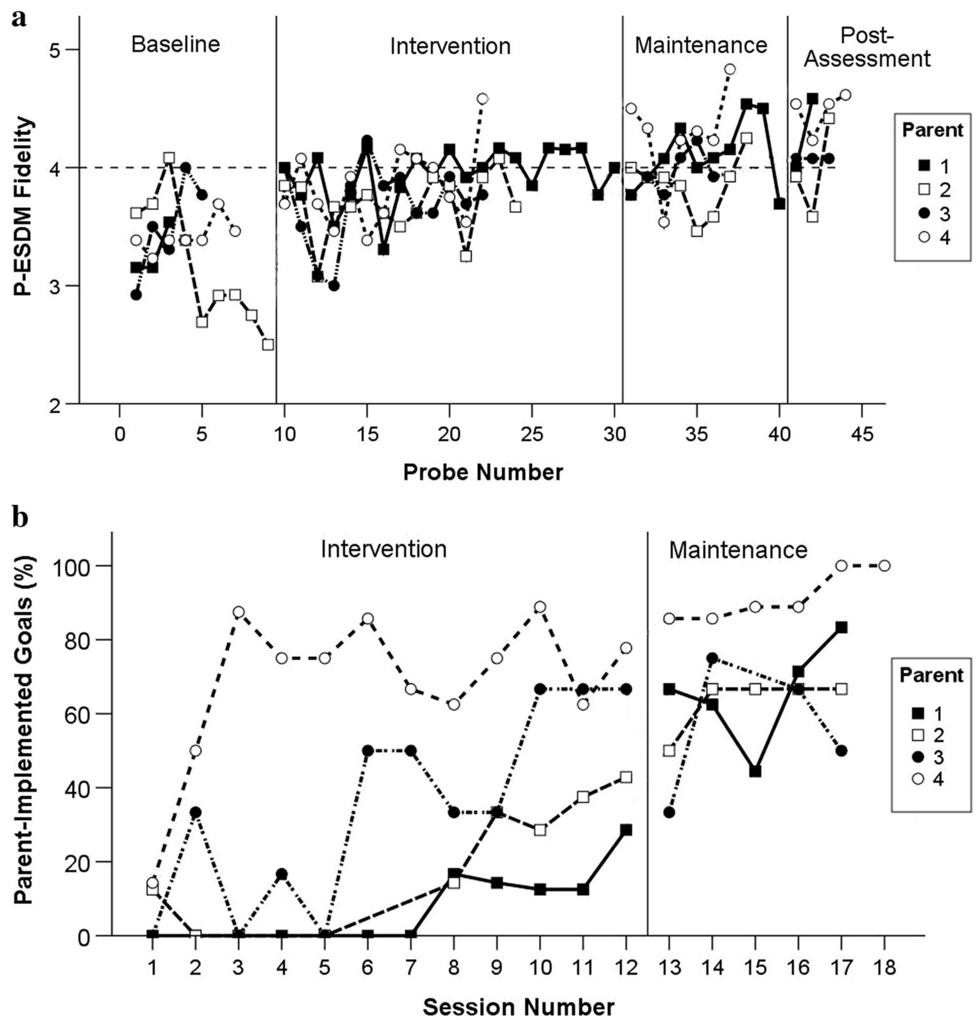

Fig. 1 **a** Parent fidelity of P-ESDM implementation across probes. **b** Percent of parent-implemented goals across sessions. Number of probes varies across parents in the study conditions

Table 3 Tau effect sizes for parent P-ESDM fidelity from baseline to maintenance and post assessment and parent-implemented goals from intervention to maintenance

Parent	Parent fidelity			Parent-implemented goals		
	Tau	SE	P	Tau	SE	P
1	0.59	0.29	0.01	0.76	0.22	0.001
2	0.72	0.22	< 0.0001	0.67	0.31	0.02
3	0.58	0.31	0.02	0.34	0.33	0.15
4	0.72	0.23	0.001	0.59	0.29	0.01

Child 2 fidelity corrected for baseline trend. Because a positive trend across intervention was expected for parent-implemented goals, these effect sizes were not corrected for trends

SD = 0.10, range 0.50–0.67). Parent 3 had some variability in implementing goals across the first few intervention sessions (mean = 0.35, SD = 0.26, range 0.00–0.43). By session 6, there was a strong trend of improvement across intervention with a flattening out across maintenance sessions (mean = 0.56, SD = 0.18, range 0.50–0.67). Parent 4 immediately improved in implementation of intervention goals (mean = 0.68, SD = 0.21, range 0.14–0.78) and demonstrated the highest rate of implemented goals out of all parents across maintenance sessions (mean = 0.92, SD = 0.7, range 0.86–1.00). Effect sizes reported in Table 3 confirmed visual analyses of the data. Parents 1, 3, and 4 significantly improved in their implementation of intervention goals from intervention to maintenance sessions. Parent 2 did not have a significant effect because levels of implementation during maintenance were similar to the highest levels achieved during intervention.

Parent Session Engagement

Parent 1 gradually increased engagement across intervention sessions (mean = 3.25, SD = 0.31, range 3.25–4.00) and maintained a high level across maintenance sessions (mean = 3.96, SD = 0.10, range 3.75–4.00). Parent 2 had a relatively lower level of engagement during all sessions (mean = 2.20, SD = 0.40, range 1.50–2.75) with no indication of improvement. Parent 3 began intervention with lower levels of engagement that steadily increased across intervention (mean = 3.27, SD = 0.59, range 2.24–4.00) and remained stable across maintenance (mean = 4.00, SD = 0.00, range 4.00–4.00). Parent 4 quickly increased engagement across intervention sessions (mean = 3.60, SD = 0.56, range 2.50–4.00) and continued high levels of engagement across maintenance sessions (mean = 3.92, SD = 0.13, range 3.75–4.00). Effect sizes reported in Table 4 confirmed visual analyses of the data. Parents 1 and 3 significantly improved engagement from intervention to maintenance sessions whereas no effect was detected for Parent 2 or for Parent 4 whose levels of engagement during maintenance were similar to the highest levels achieved during intervention.

Parent Self-Rating of Intervention Acceptance

Parent 1 rated the intervention positively throughout intervention sessions (mean = 3.90, SD = 0.22, range 3.33–4.00) with slightly higher ratings for maintenance sessions (mean = 4.53, SD = 0.52, range 4.00–5.00). Parent 2's ratings varied across intervention (mean = 3.02, SD = 0.84, range 1.00–4.00) and with consistent neutral ratings during maintenance compared to other parents (mean = 3.00, SD = 0.00, range 3.00–3.00). Parent 3's ratings gradually increased to higher acceptance across intervention (mean = 4.47, SD = 0.43, range 3.67–5.00) and remained consistently high across maintenance sessions (mean = 4.96, SD = 0.08, range 4.83–5.00). Parent 4 demonstrated a similar gradual increase across intervention (mean = 4.11, SD = 0.61, range 2.83–4.83) and maintenance sessions to favorable acceptance (mean = 4.86, SD = 0.13, range 4.57–5.00). Effect sizes reported in Table 4 confirmed visual analyses of the data. Only ratings for Parents 1 and 3 reached significance in intervention acceptance whereas there was positive but less substantial change for Parent 4 from intervention to maintenance versus less acceptance overall for Parent 2.

Coded Child Behaviors

Figure 2 demonstrates children's rate per minute of spontaneous words (top figure) and nonverbal, joint attention initiations (bottom figure). Child 1 produced less than one spontaneous word-per-minute across the initial baseline probes (mean = 0.44, SD = 0.19, range 0.28–0.65). Rates of spontaneous words continued to be generally low across intervention probes (mean = 0.46, SD = 0.55, range 0.00–1.60). Across maintenance to post-assessment, rates improved compared to baseline (mean = 1.44, SD = 1.08, range 0.00–4.29) with 11 out of the 12 probes above baseline rates. Child 2 had higher and more variable rates of words across baseline probes compared to the other participants (mean = 9.15, SD = 2.92, range 4.00–13.51). There was no indication of improvement across intervention compared to baseline probes (mean = 7.35, SD = 1.97, range 4.70–10.36). During maintenance and post-assessment, rate of spontaneous words was generally higher compared to baseline indicating some improvement (mean = 11.18, SD = 1.90, range 7.67–14.70); although still not enough to compensate for the variability demonstrated during baseline. Across baseline probes, Child 3 had mostly low rates of spontaneous words (mean = 1.22, SD = 1.65, range 0.08–4.09) and continued to be in a similar low range across intervention probes (mean = 1.97, SD = 1.07, range 0.27–3.67). There is a wider range of rates during maintenance and post-assessment (mean = 3.28, SD = 2.39, range 0.68–8.30), yet some individual probes appear similar to levels seen during baseline. Child 4 demonstrated a similar range of spontaneous words-per-minute across baseline (mean = 5.15, SD = 1.89, range 2.32–7.45) and intervention probes (mean = 4.38, SD = 1.80, range 1.06–7.87). The range increased slightly during maintenance and post-assessment (mean = 6.41, SD = 1.49, range 3.88–8.90); however only two out of 11 probes were above baseline rates. Effect sizes confirmed

Table 4 Tau effect sizes for parent self-rating of acceptance and session engagement from intervention to maintenance

Parent	Parent self-rating of intervention acceptance			Parent session engagement		
	Tau	SE	P	Tau	SE	P
1	0.67	0.26	0.01	0.51	0.30	0.04
2	−0.13	0.38	0.67	0.24	0.38	0.39
3	0.52	0.30	0.03	0.59	0.29	0.01
4	0.68	0.25	0.002	0.17	0.33	0.49

Because a positive trend across intervention was expected for parent self-rating of intervention acceptance and parent session engagement, these effect sizes were not corrected for trends

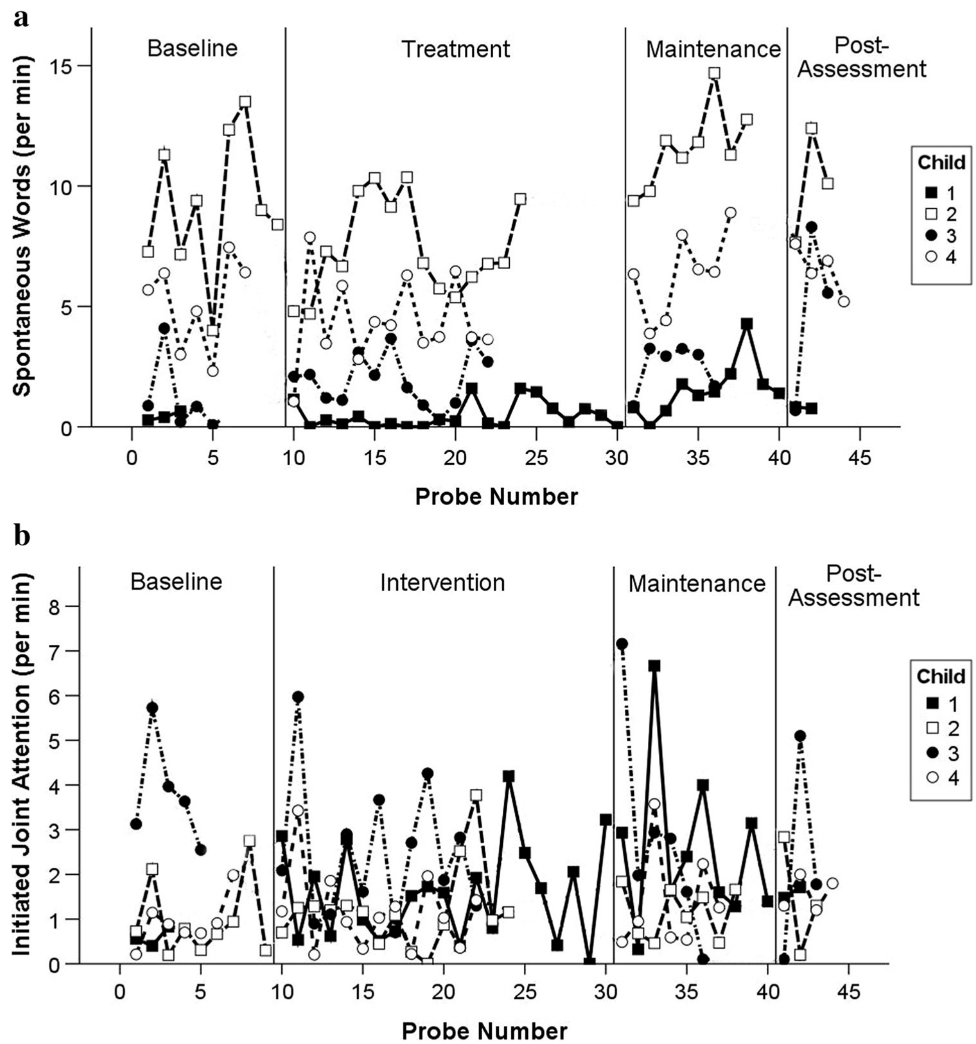

Fig. 2 **a** Child spontaneous words per minute across probes. **b**. Child joint attention initiations per minute across probes. Number of probes varies across children in the study conditions

visual analysis of trends in the data (see Table 5). Although all children had positive effect sizes indicating some increase in spontaneous words, Child 1 had the only effect size to reach statistical significance.

For rate of joint attention, Child 1 produced less than 1 joint attention initiation-per-minute across baseline (mean = 0.60, SD = 0.22, range 0.56–0.83). Rates of joint attention was more variable during intervention (mean = 1.57, SD = 1.10, range 0.00–4.20). Across maintenance and post-assessment, rate of joint attention increased (mean = 2.38, SD = 1.67, range 0.32–6.67) with only one probe falling in the range of baseline rates. Child 2 had a somewhat variable rate of joint attention across baseline (mean = 0.98, SD = 0.88, range 0.19–2.75). Variability increased across intervention (mean = 1.20, SD = 0.91, range 0.00-3.77). Rate of joint attention during maintenance and post-assessment remained similar to the rate observed across baseline (mean = 1.24, SD = 0.77, range 0.20–2.83),

Table 5 Tau effect sizes for child outcome measures from baseline to maintenance and post assessment

Child	Socially appropriate behaviors			Aberrant behaviors			Spontaneous words			Joint attention		
	Tau	SE	P	Tau	SE	P	Tau	SE	P	Tau	SE	P
1	0.62	0.37	0.09	−0.48	0.41	0.18	0.49	0.32	0.04	0.49	0.32	0.04
2	−0.76	0.38	0.10	−0.76	0.38	0.10	0.33	0.30	0.09	0.18	0.312	0.36
3	0.69	0.39	0.08	−0.46	0.31	0.48	0.39	0.35	0.11	−0.36	0.142	0.35
4	0.67	0.35	0.05	−0.23	0.49	0.65	0.30	0.32	0.16	0.27	0.322	0.21

No effects sizes were corrected for trends in baseline indicating no effect of intervention. For Child 3, baseline rates ranged between 2.55 and 5.73 (mean = 3.80, SD = 1.20). The range across the intervention phase was wider (mean = 2.45, SD = 1.51, range 0.70–5.97), with many rates below the range of observed during baseline. The variability of joint attention rates increased further across maintenance and post-assessment (mean = 2.62, SD = 2.28, range 0.10–7.16); however five out of the nine probes were below the baseline range, indicating a general trend of decreasing joint attention rates. Child 4 demonstrated similar rates of joint attention across baseline (mean = 0.93, SD = 0.55, range 0.21–1.98) and intervention (mean = 1.17, SD = 0.89, range 0.20–3.43). Across maintenance and post-assessment, the range of scores increased slightly (mean = 1.45, SD = 0.92, 0.49–3.57); however only three probes were above baseline levels. Effect sizes reported in Table 5 confirmed visual analyses of the data. There was mixed evidence for positive effects of intervention on joint attention initiations with only Child 1 demonstrating a significant increase in rates of joint attention.

Ratings of Socially Appropriate and Aberrant Behaviors

For Child 1, socially appropriate behaviors varied widely during intervention (mean = 4.00, SD = 0.69, range 2.8–5) compared to baseline ratings (mean = 3.7, SD = 0.14, range 3.6–3.8). Maintenance and post-assessment ratings appeared to be generally higher (mean = 4.17, 0.34, range 3.8–4.8), though there was still some overlap with baseline ratings. Child 2 received higher ratings for socially appropriate behavior during baseline (mean = 4.60, SD = 0.28, range 4.4–4.8) in comparison to intervention (mean = 3.92, SD = 0.23, range 3.4–4.2) and maintenance to post-assessment ratings (mean = 4.00, SD = 0.16, range 3.8–4.2). Child 3 had a small, gradual increase in ratings from baseline (mean = 3.1, SD = 0.42, range 2.8–3.4) compared to intervention (mean = 3.52, SD = 0.40, range 2.8–4) and maintenance to post-assessment (mean = 4.30, SD = 0.58, range 3.6–4.9). Child 4 had flat scores across baseline (mean = 3.00, SD = 0.00, range 3–3) and more variable scores across intervention (mean = 3.47, SD = 0.40, range 3.0–4.0). Scores across maintenance and post-assessment were higher than baseline levels (mean = 4.06, SD = 0.38, range 3.4–4.4). None of the effect sizes reported in Table 5 reached statistical significance, but they did confirm that three of the children had trends in the positive direction for socially appropriate behaviors toward parents.

For aberrant behaviors, Child 1 had steady scores across baseline probes (mean = 2.67, SD = 0.00, range 2.67–2.67) followed by variable scores across intervention (mean = 2.06, SD = 0.55, range 1.67–3.00) and maintenance to post-assessment (mean = 1.95, SD = 0.52, range 1.33–3.00). Child 2 demonstrated a slight downward trend in scores from baseline (mean = 2.50, SD = 0.24, range 2.33–2.67) compared to intervention (mean 2.43, SD = 0.55, range 1.33–3.00) and maintenance to post-assessment (mean = 1.58, SD = 0.32, 1.33–2.00). Although Child 3's scores were stable across baseline probes (mean = 2.00, SD = 0.00, 2.00–2.00), scores were variable with no discernable pattern of change across intervention (mean = 1.61, SD = 0.37, 1.00–2.33) and maintenance to post-assessment (mean = 1.60, SD = 0.43, 1.33–2.33). Child 4 had similar variable scores across baseline (mean = 1.67, SD = 0.47, 2.00–1.33), intervention (mean = 2.12, SD = 0.58, 1.00–3.00), and maintenance to post-assessment (mean = 1.62, SD = 0.52, range 1.00–2.33). No effect sizes in Table 5 reached significance.

Standardized Child Assessments

Children 1, 2, and 3 each had an overall intervention duration (including intervention and maintenance phases) of 6 months between baseline and follow up assessments, and child 4 had a 9-month overall intervention period due to the therapist's relocation. These time periods can be compared to the number of developmental months gained on the Vineland and MSEL (see Table 6). On the Vineland, by parent report, the three boys (children 1, 3, and 4) in the study made, on average, 5–7 months of developmental gains compared to the 6–9 months elapsed between assessments, and the girl (child 2) made on average 14 months progress across 6 months' time. This is reflected in the increase in her adaptive behavior composite (89–98). The boys either remained consistent or had a modest drop in composite scores despite making gains. There was no clear pattern of change in Vineland domains across children.

On the MSEL, perhaps a more objective, but less ecologically valid measure of developmental progress, the three boys remained relatively stable with regard to their overall Early Learning Composite reflecting a typical, not delayed rate of cognitive growth, whereas the girl showed a 12-point gain, largely driven by a nearly 2-year developmental leap in Visual Reception. An exception to this pattern was child 3, whose 4-point drop on the composite appears to be due to an 8-month loss in Receptive Language. This however is likely due to variation in attention or mood across assessments rather than representing a true loss of language. As such, the drop in standard score should be interpreted cautiously.

On the MacArthur-Bates Communication Inventories, all four parents reported gains, especially in their child's use and understanding of gesture production. As these are not standard scores, it is not possible to make clear interpretations regarding the significance of the increased language, but the gains of children 1 and 3 appear quite substantial.

Table 6 Pre and post standardized assessment results

ADOS total score/classification	Child 1			Child 2			Child 3			Child 4		
	11 ASD			5 No ASD			13 ASD			10 ASD		
	Pre	Post	Δ	Pre	Post	Δ	Pre	Post	Δ	Pre	Post	Δ
Chronological age	2:5	2:11	6	3:6	4:0	6	2:11	3:5	6	2:3	3:0	9
VABS II (age equivalent)												
Communication	2:4	3:1	7	2:3	3:5	14	1:4	2:5	13	2:0	2:7	7
Daily living skills	–	1:5	–	2:10	4:2	16	1:3	2:0	9	2:6	3:0	6
Socialization	–	1:8	–	3:4	4:7	13	1:2	1:10	8	2:2	3:0	10
Motor skills	2:3	2:8	5	3:7	4:6	11	2:2	2:9	7	1:10	2:4	6
Adaptive behavior composite	–	79		89	98		72	73		95	87	
Mullen (age equivalent)												
Visual reception	1:1	2:2	13	2:8	4:6	22	1:9	2:5	8	1:9	2:2	5
Fine motor	1:3	1:10	7	3:0	3:4	4	1:9	2:2	5	1:8	1:8	0
Receptive language	1:8	2:1	5	2:7	3:0	5	2:4	1:8	-8	1:11	2:7	8
Expressive language	0:7	1:4	9	2:3	2:9	6	1:0	1:10	10	1:11	2:5	6
Early learning composite	54	57		70	82		55	51		70	70	
MCDI												
Phrases understood	25	26		13	27		13	19		25	28	
Comprehension	204	280		302	394		122	194		271	380	
Production	1	98		248	394		5	54		260	380	
Total gestures used	33	42		57	60		38	46		55	58	

ADOS Autism Diagnostic Observation Schedule; *VABS II* Vineland Adaptive Behavior Scales, Second Edition; *Mullen* Mullen Scales of Early Learning; *MCDI* MacArthur-Bates Communicative Development Inventories; – score not attainable

Without a control group for comparison though, it is not possible to confidently attribute gains to the intervention.

Discussion

Empirically-supported behavioral intervention methods are still lacking for children with FXS with or without ASD let alone an approach to include parents in this interactive process to promote early critical developmental skills. To our knowledge, this is the first study to describe effects of a well-standardized parent-coaching model for children with ASD and used with children who have a known genetic disorder associated with autism. Since the scope of this study was preliminary, we attempted to pilot test the initial feasibility and acceptability of coaching parents to use the P-ESDM with their child. A subsequent objective was to obtain findings to empirically inform future research using a larger sample in a controlled clinical trial.

Our first research question examined parent implementation with the P-ESDM intervention through fidelity attainment and intervention goals taught to children. We focused on these two measures as gauges of parent learning since the causal model attributes treatment-related child change to improvements in the frequency, sensitivity, and responsivity of parent–child interactions. Through increased parent fidelity performance, parents were more likely to follow their child's interests and supported their play actions to build fun activities and a shared focus as coaching and practice with the intervention progressed. Within this meaningful social context, three out of the four parents also provided more and diverse opportunities for learning measured by the frequency to which they practiced defined intervention goals for interaction, play, emotional regulation, and communication.

Of particular importance to social interactions with children with FXS was parents' sensitivity to the P-ESDM strategies for optimizing their arousal and affective engagement. Research stresses the importance of social engagement and enhanced positive affect to learning (Kasari et al. 1990; Messinger et al. 2001; Hohenberger 2011) because it helps children attend to parents' faces and expressions and process what they are saying, sharing, and showing. Children then have more opportunities to make sense of parents' linguistic, social, affective, and emotional information. However, FXS can negatively heighten emotional and attentional domains through exaggerated sympathetic nervous system and limbic system activity in response to social, cognitive and sensory challenges (Cordeiro et al. 2011; Farzin et al. 2006; Hessl et al. 2006, 2002; Miller et al. 1999). With the present sample, coaching sometimes involved parents increasing the child's arousal to enhance attention and positive affect during more physical and anticipatory social interactions (e.g., offering a game of tickles or hide-and-seek). Other times it involved parents' selection of calmer, soothing activities (e.g., reading a book, listening to music, drinking juice) and adjusting their tone and pace to dampen and regulate the child's arousal. Parents' attention to their child's regulation, as well as use and alternation of therapeutic strategies seemed to stabilize their child's affect and arousal systems for longer engagement and increased social orienting as evidenced by gradual lessening in common behaviors associated with FXS (i.e., anxious, fearful, withdrawn, panicked). Instead, children displayed pleasure and enjoyment from parental sensitivity and modulation of arousal techniques with the goal of making social engagement with the parent an intrinsic part of the reward and a reason to continue coming back to the parent and activity.

Three out of the four parents reported positive ratings with the P-ESDM intervention and coaching experience. They reported increased confidence and capability blending the P-ESDM strategies into everyday activities with their child. They noted specific examples of how the intervention generalized to their daily life (i.e., "he brings me toys to play with him," "he wants to sing with me in the car") and to other family members or play partners (i.e.., "I showed his grandparents how to work on his learning goals" "she said hi to another child"). In addition, raters observed their increased communication with the coach (e.g., sharing personal experiences, asking questions, elaborating on what the coach said), comfortable body language (e.g., eye contact, open posture, smiling), and understanding of the intervention content and strategies (e.g., generating learning moments or solutions without the coach's assistance). Parent 2 may have found the intervention program less helpful and specific to her goals that concentrated more on self-help skills for her child, such as potty-training, sleeping in her own bed, and dressing independently, than the social-communication focus of the P-ESDM. Her evaluation of the Likert-based rating statements may have reflected her dissatisfaction with the intervention content and/or participating in center-based sessions where it was more difficult to successfully support progess with such skills. While we still have more to understand about how parents engage with, respond to, and perceive the P-ESDM intervention, the methods are important to continue studying because of the potential value children attach to shared engagement necessary for learning, social reciprocity and emotion regulation. We know from studies of typical brain function that emotionally salient events are attended to more readily, elicit greater brain activity, and are more likely to be remembered than when not provided (Markovic et al. 2014). We want interactions from children's perspectives to be valued as more rewarding with parents included than excluded so that memorable exchanges lead to repeated engagement and thus learning to occur. These outcomes are particularly important given the difficulty and importance of skill generalization for children with developmental disabilities and the concern about parent-mediated intervention and their potential for increasing parent stress.

We then examined the effects of parent-mediated intervention on child progress. Individual change and effect sizes on spontaneous communication and initiated joint attention behaviors varied across children, making it difficult to draw firm conclusions about child change. We see from this and other autism parent-mediated treatment (e.g., Carter et al. 2011; Rogers et al. 2012b) that the abbreviated and low-intensity nature of the intervention and focus on parent mediation (as opposed to therapist implementation) may require a greater dosage for parent effects to "trickle" downward for child change (Green et al. 2015; Rogers et al. 2018). The proxy child behaviors targeted in the study also represent complex skills and core deficits challenging to change. For some children, specific attention and skill building may be necessary to see effects emerge from abbreviated parent-mediated models. The work of Connie Kasari and colleagues has shown strong, significant gains in children's joint attention, language, and play skills from their manualized, skill-focused curriculum (i.e., Joint Attention Symbolic Play Engagement and Regulation, JASPER) and when delivered by parents (Kasari et al. 2010, 2014). The P-ESDM also includes specific developmental strategies to help parents facilitate joint attention growth in children; however, its curriculum is broader and parents may need more time and practice to get through and feel competent using all content. As we refine a model of behavioral intervention for FXS and ASD, considerations include whether we need to approach specific developmental areas, such as joint attention, with a greater emphasis on skill teaching or reallocate coaching to certain fidelity behaviors that have a stronger pivotal effect on other interactive behaviors helpful to child learning and developmental outcomes. Other FXS deficit areas, such as attention and hyperactivity/impulsivity, may need to be targeted as goals in treatment as well to lead to robust developmental gains.

Gains occurred on standardized composite scores of adaptive behavior, visual reception, fine motor, expressive, and for all but one child, receptive language suggesting that three out of four children made comparable developmental ground relative to their same-aged typical peers. In some domains, children's gains exceeded normative expectations (e.g., gained more months in age equivalence than chronological months of growth). Parents also reported an increase in perceptions of children's understanding and use of communicative gestures and words.

Overall results suggest both the potential promise and limitations of this study. Strengths include a manualized autism empirically-supported intervention program with defined teaching content, coaching tools, and fidelity systems to measure parent and coaching implementation. This is important for quality control work with families, as well as replication to occur in future studies. Parents gained and enjoyed using interactive skills important to child developmental needs not easy to change and from coders uninvolved with the intervention. Although general maturation or the amount and influence of other parenting interventions cannot be ruled out, the lack of significant IQ gains that occur in untreated children with ASD in early childhood research (Lord et al. 2005; Sigman et al. 1999) suggest that it is not unreasonable to believe that intervention facilities positive change. These are some of the first data on children with a known genetic disorder in response to an autism standardized behavioral, parent-mediated approach.

Nonetheless, we acknowledge the clear shortcomings (i.e., very small sample size, no comparison group, limited baseline data, variability in parent–child dyad outcomes, low threshold of kappa levels) that cannot give definitive conclusions about the efficacy of this intervention or how the short-term nature of a parent-mediated intervention model may support the range of attention-deficit/hyperactivity, sensory hypersensitivity, repetitiveness, and anxiety/hyperarousal features of FXS and ASD characteristics without further testing (Boyle and Kaufmann 2010; Hagerman et al. 2009; Talisa et al. 2014). Parents who enrolled in this study also demonstrated considerable motivation for their children's treatment and were advantaged in ways (i.e., education, social support) that may differ for how families in the broader population respond to this approach. Further the level of clinical skill and adherence to the treatment delivery in this study from a co-developer of the model is likely higher than would be found in community settings. A community-implementation study of P-ESDM is underway to determine its effectiveness in everyday community settings and with lower-resourced families typically excluded from clinical treatment opportunities. Finally a larger replication of in-person versus telehealth parent-mediated intervention that was beyond the scope of this study could examine the learning advantages versus limitations on parent–child outcomes. Utilizing distance technology is of particular importance when conducting research with low incidence populations, such as FXS, and may increase service options to families who routine face limited access and/or difficulty traveling to a clinic with a child with a developmental disability (Bailey et al. 2003; Statham et al. 2011). For some families, the flexibility of telehealth to connect from their home may provide a unique opportunity for the support and practice necessary for a low-dosage parent-training model to produce noticeable behavioral changes (McDuffie et al. 2016) whereas other families may require more intensity than can be effective with telehealth (Vismara et al. 2016).

In closing, this study is the first to our knowledge to examine the preliminary efficacy and acceptability of a well-standardized parent-coaching model targeting social and communicative development for young children with a developmental disorder other than ASD. We cannot determine causality and our results are not definitive. The pilot study does however support further research on the potential impact of this parent-mediated intervention during a chronologically younger developmental period when learning is critical and parents can provide salient social and linguistic information for brain development. Future understanding is imperative given there are few behavioral interventions that have been empirically tested for families of young children with FXS and ASD.

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Author Contributions LAV designed and implemented the study, co-assisted in the analyses of data, wrote the bulk of the manuscript, and addressed the reviewers' comments and revisions made to the manuscript. CEBM conducted the statistical analyses, lead the analysis of the data, wrote the results section of the manuscript, and responded with revisions to the statistical analyses, data, and results section of the manuscript. RS participated in the coding of data. DH provided clinical guidance on the implementation of the study, developed and made changes per reviewers' requests to the figures representing the data, and assisted with manuscript development and revisions to the manuscript addressing the reviewers' comments.

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Compliance with Ethical Standards

Conflict of interest Potential conflicts of interest for Laurie A. Vismara involve royalties from publications and honoraria received from lectures related to the parent-delivered Early Start Denver Model. David Hessl serves on the Scientific Advisory Board and the Clinical Trials Committee of the NFXF.

Ethical Approval All procedures performed in this study involving human participants were in accordance with the ethical standards of the institutional research committees and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed Consent Informed consent was obtained from parents or legal guardians of all individual participants for whom identifying information is included in this article. No adverse effects of the study occurred.

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