Early Language Intervention Using Distance Video-Teleconferencing: A Pilot Study of Young Boys With Fragile X Syndrome and Their Mothers

Andrea McDuffie, Ashley Oakes, Wendy Machalicek, Monica Ma, Lauren Bullard, Sarah Nelson, and Leonard Abbeduto

Purpose: This study examined the effects of a naturalistic parent-implemented language intervention on the use of verbally responsive language by mothers of 6 young boys with fragile X syndrome. The intervention included parent education sessions and clinician coaching delivered onsite and by distance video-teleconferencing.

Method: A single-case multiple baseline across participants was used to examine intervention effects on maternal use of language support strategies. A nonparametric analysis was used to evaluate the relative effectiveness of onsite compared with distance coaching sessions.

Results: Mothers increased their use of utterances that followed into their child's focus of attention and prompted child communication acts. Intervention effects were not observed for maternal contingent responses to child communication, possibly due to the limited number of spontaneous communication acts children produced. Children showed moderate increases in the use of prompted communication acts, whereas intervention effects on spontaneous communication acts were more modest and variable. Comparable increases in maternal strategy use were observed during onsite and distance sessions.

Conclusions: No previous study has examined a distance-delivered parent-implemented language intervention for young boys with fragile X syndrome. Mothers were able to increase their use of verbally responsive language. Intervention efficacy might be enhanced by incorporating an augmentative and alternative communication device for some children and a more concerted focus on increasing the frequency of child communication acts. Findings provide preliminary support for the efficacy of the distance delivery format.

Responsive verbal language input from parents has been found to make an important contribution to early language development for typically developing children (e.g., Tamis-LeMonda & Bornstein, 2002) as well as for children at risk for or experiencing developmental delays (e.g., McDuffie & Yoder, 2010; Siller & Sigman, 2002; Warren, Brady, Sterling, Fleming, & Marquis, 2010). Responsive verbal language follows the child's lead, is affectively positive, and is provided promptly and contingently as the parent talks about their child's focus of attention or responds to their child's nonverbal or verbal communication acts (Bornstein, Tamis-LeMonda, & Haynes, 1999). Such parental input facilitates early language learning by making the correspondence between words and their referents explicit and by minimizing the cognitive and attentional resources that children must devote to language learning. Increases in parent verbal responsiveness are frequently targeted in naturalistic language intervention programs for young children with intellectual disabilities (Fey et al., 2006; Yoder & Warren, 2002) as well as those with autism spectrum disorders (ASD; Green et al., 2010; Ingersoll & Gergans, 2007; Venker, McDuffie, Ellis Weismer, & Abbeduto, 2012; Yoder & Stone, 2006). There are, however, no published studies that have examined the efficacy of such interventions for mothers of young boys affected by fragile X syndrome (FXS), the leading inherited form of intellectual disability. This is an important population to consider given that the behavioral characteristics of young boys

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with FXS and their mothers are likely to contribute to interactions that are less than optimal for language learning.

**FXS and Associated Disorders**

FXS results from the mutation of a single gene (FMR1) on the X chromosome (Brown, 2002). In the full mutation of the FMR1 gene, a repetitive sequence of trinucleotides (i.e., the CGG repeats) expands to 200 or more from its unaffected level of 54 or fewer repeats. This expansion leads to methylation and transcripational silencing of the gene, reducing or completely eliminating its protein product, FMRP (Oostra & Willemsen, 2009). FMRP is critical for experience-dependent learning through its effects on the maturation and pruning of synapses (Bhakar, Dölen, & Bear, 2012). Individuals who have 55 to 200 CGG repeats carry the FMR1 premutation and can have reduced levels of FMRP as well as increased—and perhaps toxic—levels of FMR1 messenger ribonucleic acid (Tassone & Hagerman, 2003). Prevalence estimates are one in 3,500 boys and men and one in 6,000 to 8,000 girls and women for the full mutation (Coffee et al., 2009) and one in 468 boys and men and one in 151 girls and women for the premutation (Seltzer, Barker, et al., 2012). Boys and men with the FMR1 full mutation tend to be more affected than girls and women given the protective presence of an active healthy X chromosome in girls and women. Regardless of sex, however, individuals with the FMR1 premutation or full mutation are at higher risk for suboptimal developmental outcomes (Jacquemont, Hagerman, Hagerman, & Leehey, 2007).

**Boys and Men With FXS**

Virtually all boys and men with the FMR1 full mutation have cognitive delays, with the vast majority having IQs between 40 and 55 (Hessl et al., 2009). In addition to cognitive delays, the behavioral phenotype, or profile of behaviors that is characteristically observed (J. Harris, 2002) in boys and men with this genetic disorder, includes significant delays in language development (J. Roberts, Mirrett, Anderson, Burchinal, & Neebe, 2002), even relative to what would be expected on the basis of nonverbal developmental level (Abbeduto, Brady, & Kover, 2007). The majority of boys and men with FXS also display hyperactivity and attentional difficulties (Scerif, Longhi, Cole, Karmiloff-Smith, & Cornish, 2012), anxiety and withdrawal (Cordeiro, Ballinger, Hagerman, & Hessl, 2011), and repetitive (Hall, Lightbody, Hirt, Rezvani, & Reiss, 2010) and challenging behaviors, including aggression and self-injury (Leigh, Hagerman, & Hessl, 2013; Symons, Clark, Hatton, Skinner, & Bailey, 2003). Challenging behaviors emerge early in toddlerhood for boys with FXS (Symons et al., 2003; Symons, Byers, Raspa, Bishop, & Bailey, 2010) and are frequent responses to changes in routines, task demands, and the desire for preferred items (Machalicek et al., 2014). Up to 60% of boys and men with FXS display behaviors that are frequent and severe enough to warrant a comorbid diagnosis of an ASD (Bailey et al., 1998; Demark, Feldman, & Holden, 2003; Hall et al., 2010; S. Harris et al., 2008).

**Mothers of Children With FXS**

As carriers of the FMR1 premutation or full mutation (Nolin et al., 1996), the psychological well-being of mothers of children with FXS may be affected by the mothers’ own biology, including an increased risk of depression and anxiety in premutation mothers, particularly in the context of parenting stress and stressful life events (Seltzer, Barker, et al., 2012; Wheeler et al., 2014). In fact, there is evidence that these women are more likely to exhibit symptoms of anxiety and depression than mothers of children with other disabilities (Abbeduto et al., 2004; Lewis et al., 2006). Moreover, even women with the premutation who do not have affected children have higher rates of mental health problems compared with the general public (Bailey, Sideris, Roberts, & Hatton, 2008; Franke et al., 1996; J. Roberts et al., 2009). It should be noted, however, that the psychiatric symptoms associated with premutation status affect only a subset of carrier mothers. In particular, recent studies have examined various aspects of the premutation phenotype, including repeat length and activation ratio (i.e., the proportion of cells that produce normal levels of FMRP), and have suggested that the association between these biomarkers and psychiatric symptoms may be nonlinear; that is, there may be increased vulnerability in girls and women with midrange repeats (Hartley et al., 2012; Loesch et al., 2014; J. Roberts et al., 2009). In the present context, higher levels of maternal stress and poorer psychological well-being are of concern because these characteristics may decrease maternal verbal responsivity and thereby increase children’s risk for language learning problems (e.g., Landry, Smith, & Swank, 2006). The delivery of a parent-mediated intervention may encourage more positive parent–child interactions and may teach mothers ways to support their children’s language development. The indirect effects of interventions that address patterns of parent–child interactions have yet to be examined for children with FXS (Tonnsen, Cornish, Wheeler, & Roberts, 2014).

**Verbal Responsivity and Language Development in FXS**

It is likely that the behavioral phenotype of boys and men with FXS contributes to their language delays in several ways. Repetitive behaviors, hyperarousal, and inattention may restrict interest in actively exploring the environment and in maintaining productive engagement with objects (McDuffie et al., 2015). Engaging in repetitive behaviors can occupy children’s attention such that they do not notice, explore, and learn about the functions of common objects in their environments. Hyperarousal and inattention may disrupt sustained object engagement and prevent children from learning about the properties of objects and how objects can be used or combined. Escape-maintained challenging behaviors (Machalicek et al., 2014) also may interfere with meaningful participation in daily routines that support language learning. Last, child challenging behaviors may lead mothers to be directive rather than verbally responsive (Brady, Warren, & Sterling, 2009).
There is evidence from longitudinal correlational studies of parent-child interaction, however, that children with FXS do have more positive language outcomes if their mothers are verbally responsive (Warren et al., 2010) and if increased levels of verbal responsivity are sustained across childhood (Brady, Warren, Fleming, Keller, & Sterling, 2014). These and other descriptive studies focusing on other groups of children with intellectual and developmental disabilities (e.g., ASD; Haebig, McDuffie, & Ellis Weisner, 2013; McDuffie & Yoder, 2010; Siller & Sigman, 2002) provide a rationale for providing an intervention that targets increases in maternal verbal responsiveness as a means of supporting language development in young children. In such an intervention, parents would mediate the effect of the intervention on child communication and language outcomes.

Parent-Implemented Language Interventions

Relative to clinician-implemented intervention approaches, there are several advantages to training parents so that they can, in effect, serve as the interventionist for their child. Parents can embed language teaching into naturally occurring routines and activities, thereby providing numerous opportunities for turn taking and communicative interactions throughout the day (Kashinath, Woods, & Goldstein, 2006). Child learning occurs in naturalistic contexts with naturally occurring reinforcers and thus is more likely to generalize to new situations and to be maintained over time (Kaiser, Hancock, & Hester, 1998). Parent-implemented programs provide a higher intensity of exposure to intervention content than is possible in clinician-implemented treatments (Ingersoll & Gergans, 2007). Moreover, numerous studies have demonstrated that maternal verbal responsiveness can be increased within the context of an intervention that includes a parent education component (e.g., Bottema-Beutel, Yoder, Hochman, & Watson, 2014; Green et al., 2010; Kasari, Gulsrud, Wong, Kwon, & Locke, 2010; Kashinath et al., 2006; Yoder & Stone 2006), provided that parents receive coaching as part of their participation in the intervention program (Kaiser, Hancock, & Trent, 2007).

Although very few published studies have described the implementation of such an intervention approach for young boys with FXS, Brady et al. (2009) reported on a pilot study in which More Than Words, a naturalistic, parent-mediated language intervention program developed by the Hanen Center (Sussman, 1999), was individually delivered to mothers of four young boys with FXS. Three of the four mothers increased the proportion of responsive interactions and decreased their proportion of directive interactions with their children. Likewise, children of these three mothers showed concomitant increases in spoken vocabulary and rates of communicative initiations. These results, in addition to the results of other studies of parent-implemented interventions, suggest that this may be a viable intervention approach for young children with FXS.

Telepractice: Intervention Delivery at a Distance

The American Speech-Language-Hearing Association defines telepractice as the distance delivery of professional services by a speech-language pathologist (SLP), accomplished by linking the clinician to a client or professional using technology-based software applications. There is still a relative dearth of research specifically examining the efficacy of distance delivery for the provision of pediatric language intervention services by SLTs. To date, most telepractice services implemented by SLTs have involved the assessment and treatment of voice disorders, stuttering, and adult neurogenic communication disorders (Mashima & Doarn, 2008; Theodoros, 2011). In addition, there are numerous reports on the use of telepractice for the assessment and treatment of individuals with ASD (Boisvert, Lang, Andrianopoulos, & Boscardin, 2010). Telepractice has been used to deliver a variety of services to individuals with this diagnosis, including training teachers to implement preference assessments and functional behavior assessments (Barretto, Wacker, Harding, Lee, & Berg, 2006; Machalicek et al., 2009) as well as training teachers (Gibson, Pennington, Stenhoff, & Hopper, 2010; Vismara, Young, Stahmer, Griffith, & Rogers, 2009) and parents to implement behavioral interventions (McDuffie et al., 2013; Vismara, McCormick, Young, Nadhan, & Monlux, 2013; Vismara, Young, & Rogers, 2012; Wainer & Ingersoll, 2011).

Relative to face-to-face delivery of early intervention services in the clinic or at home, one of the foremost advantages of Internet-based delivery approaches is the potential to reach all individuals for whom an intervention is intended (Baggett et al., 2010). Thus, the continued development of telepractice has been driven, in part, by its cost efficiency and the provision of equitable access to services (Theodoros, 2011). In addition, telepractice can accommodate individual scheduling needs and may provide improved fidelity of implementation (Baggett et al., 2010). Last, it has been suggested that telepractice may optimize the timing, intensity, and sequencing of intervention content, leading to improved functional outcomes for clients.

A recent meta-analysis of parent-implemented language interventions for children between 18 and 60 months of age (M. Roberts & Kaiser, 2011) reported that relatively few early intervention activities actually were conducted within family homes, although this represents the setting most frequently identified as the natural environment for families of young children (Hebbler et al., 2007). The use of distance video-teleconferencing offers the potential benefit of supporting parents’ use of development-enhancing routines and activities within the home—an intervention setting that is recommended by many professionals (e.g., Wilcox & Woods, 2011).

At the present time, there is only one published study that examined the preliminary effectiveness of a parent-implemented early language intervention delivered by SLTs using distance telepractice (McDuffie et al., 2013). The McDuffie et al. (2013) study examined the effects of a naturalistic language intervention on the use of three language
support strategies by mothers of eight preschoolers with ASD. This intervention lasted 16 weeks and consisted of four monthly parent education sessions held onsite at a university-based clinic. Each parent education session was paired with face-to-face coaching of a play-based parent–child interaction. Twelve weekly distance coaching sessions also were implemented via video-teleconferencing using a laptop computer and Skype (http://www.skype.com/en). Parents increased their use of comments that described their child’s current focus of attention and increased their contingent verbal responses to child nonverbal and verbal communication acts. Parents also increased the frequency with which they used choices, environmental arrangement, and time delay to prompt child communication acts, and children increased their frequency of prompted communication acts. Increases in parent strategy use were observed at equivalent levels during both onsite and distance coaching sessions. The current study utilized the same experimental design and intervention model (i.e., four parent education sessions, four face-to-face clinician coaching sessions, and 12 distance coaching sessions) and targeted the same parent strategies (follow-in commenting, prompting child communication, and contingent responses to child communication acts) as the McDuffie et al. (2013) study; however, the present study focused on boys with the FXS full mutation and their mothers.

Research Questions

The goals of the current study were to (a) gather preliminary data on the efficacy of a naturalistic language intervention for young boys with FXS designed to increase maternal verbal responsiveness and (b) evaluate the effectiveness of combining face-to-face parent education and coaching with parent coaching provided at a distance using distance video-teleconferencing. The following research questions were addressed:

1. Does the intervention lead to increases in maternal use of follow-in commenting?
2. Does the intervention lead to increases in maternal use of indirect communication prompts and child-prompted communication?
3. Does the intervention lead to increases in child spontaneous communication acts?
4. Does the intervention lead to increases in maternal use of contingent verbal responses to child communication acts?
5. Does parent use of targeted strategies during distance coaching sessions equal or exceed strategy use during onsite coaching sessions?

Method

Participants and Setting

Six boys and their biological mothers participated in the study. Families were recruited from a Listserv of families affected by FXS. Children met the following eligibility criteria: (a) a confirmed diagnosis of full-mutation FXS, (b) between 2 and 6 years of age, (c) no two-word spoken utterances according to maternal report, (d) English as the primary language spoken in the home, and (d) no uncorrected sensory or motor impairments severe enough to preclude processing and responding to verbal language input. One child was taking a daily dose of a prescription medication for attention-deficit/hyperactivity disorder, and another child was taking a daily dose of a prescription medication for anxiety. Five dyads were from the United States, and one dyad was from Canada. These six participants were part of a cohort of 12 young boys with FXS who provided the samples for three other publications (Machalicek et al., 2014; McDuffie et al., 2015; Oakes, Ma, McDuffie, Machalicek, & Abbeduto, 2014).

A battery of standardized tests and informant report measures was administered to each dyad during pre- and posttreatment visits conducted at a university-based research center in the western United States. Informed consent was obtained from all mothers, and all intervention procedures were approved by the institutional review board. Characteristics of each dyad at the pretreatment visit are presented in Table 1.

Video-Teleconferencing Equipment

Equipment loaned to each family consisted of a 13.3-in., 2.4-GHz, 250-GB hard drive/SuperDrive MacBook laptop computer (Apple, Cupertino, CA) and an external Logitech (Romanel-sur-Morges, Switzerland) QuickCam Pro 9000 camera. The camera was connected to the laptop and positioned so that the clinician could view the area in the home where the session took place. The clinician used the same model of laptop and initiated video calls using the built-in iSight web camera and Skype software. Distance sessions were captured using eCamm Call Recorder software (http://www.ecamm.com). Computers were connected to broadband Internet by Ethernet cable or wireless connection. Confidentiality of data transmission was secured through 128-bit advanced encryption.

Design of the Intervention Program

A nonconcurrent multiple-baseline design (Christ, 2007; Harvey, May, & Kennedy, 2004; Watson & Workman, 1981) across participants was used to assess the efficacy of the intervention during onsite and distance coaching sessions. In this single-case design, the investigator predetermines the lengths of each of several baseline phases to be implemented at different points in time (Watson & Workman, 1981). Conducting this study using concurrent baselines across tiers (i.e., participants) would have required that at least three families travel to the research clinic during the same time frame, which would have been difficult to accommodate in terms of lab staffing. As each participating dyad entered the study, they were randomly assigned to a baseline length until all predetermined baseline lengths were utilized. In the current study, we randomly assigned two
dyads each to baseline lengths of five, eight, and 11 sessions, thus forming two groups of three participants. Each group of three participants represented a separate multiple-baseline design and consisted of a participant who received five, eight, or 11 baseline sessions. Although baseline data were not collected concurrently, changes in targeted behavior during treatment can be considered to be functionally related to the treatment because they occurred in conjunction with treatment procedures that were implemented at randomly assigned points in time (Watson & Workman, 1981).

For research questions 1 through 4, visual analysis was used to examine changes in each dependent variable. In addition, effect sizes were calculated using nonoverlap of all pairs (NAP; Parker & Vannest, 2009), an index summarizing the degree of nonoverlap between baseline-phase data and intervention-phase data. NAP represents the percentage of data that improve across phases (Parker, Vannest, & Davis, 2011). NAP statistically is equivalent to the area under the curve in Receiver Operating Characteristic analysis or a Mann–Whitney U test in nonparametric analysis (Parker & Vannest, 2009). In calculating NAP, the denominator represents the total number of comparisons between baseline data points and intervention data points ($N_b \times N_i$). The numerator is computed by comparing each baseline data point with each intervention data point and then calculating all overlapping pairs between baseline and intervention, with ties (e.g., baseline = 5, intervention = 5) receiving .5 point and comparisons in which the value of a baseline data point exceeds the value of an intervention data point receiving 1 point. The sum of points generated from overlapping pairs is subtracted from and then divided by the product term: $[(N_b \times N_i) - \text{Sum overlapping pairs}] / (N_b \times N_i)$. The value of NAP ranges from .50 to 1.00. At a value of NAP = .50, there is a 50% chance that scores from the treatment phase cannot be differentiated from scores from the intervention phase. According to Parker and Vannest (2009), NAP values under .65 represent weak intervention effects, values between .66 and .92 represent moderate effects, and values between .93 and 1.00 represent strong effects.

Nonparametric statistics were used to address research question 5. In general, changes in maternal behavior were considered to represent proximal changes attributable to the intervention, whereas changes in child behavior were considered to represent more distal changes. Standardized testing was not repeated after the conclusion of the intervention. Given the developmental level of the children, it would not be likely that they would make substantial changes in standardized assessments over a period of only 16 weeks.

### Structure of the Intervention Program

#### Baseline Sessions

Dyads 1, 2, and 3 were compared with one another in one multiple-baseline design, whereas Dyads 4, 5, and 6 were compared with one another in an additional multiple-baseline design. Dyads 1 and 4 were the first to enter baseline, with an additional pair of dyads entering the study each week. Each baseline session lasted 10 min. Baseline sessions for Dyads 1 and 4 were collected during a single day. Baseline sessions for Dyads 2, 3, 5, and 6 were collected over two sequential days. For each baseline session, the dyad was provided with three developmentally appropriate toys, and the mother was instructed to play with her child as she usually would. A new toy set was provided for each baseline session, and breaks were taken to complete other testing or as needed. On average, breaks between baseline sessions were 30 min. Participation in the intervention was uniformly high; mothers completed all onsite visits (explained below), and only one distance session was not completed across all six dyads.

#### Parent Education Lessons

The intervention provided an onsite parent education lesson during each visit to the clinic for a total of four parent education lessons. Each parent education lesson lasted approximately 90 min and was implemented by a licensed SLP (the first author) and a masters-level speech-language

### Table 1. Characteristics of participating dyads at preintervention.

<table>
<thead>
<tr>
<th>Variable</th>
<th>Dyad 1</th>
<th>Dyad 2</th>
<th>Dyad 3</th>
<th>Dyad 4</th>
<th>Dyad 5</th>
<th>Dyad 6</th>
</tr>
</thead>
<tbody>
<tr>
<td>Child</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Chronological age (months)</td>
<td>39</td>
<td>35</td>
<td>40</td>
<td>27</td>
<td>40</td>
<td>43</td>
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<tr>
<td>Words understood(^a)</td>
<td>129</td>
<td>325</td>
<td>254</td>
<td>336</td>
<td>179</td>
<td>233</td>
</tr>
<tr>
<td>Words produced(^a)</td>
<td>1</td>
<td>71</td>
<td>0</td>
<td>33</td>
<td>2</td>
<td>40</td>
</tr>
<tr>
<td>Comprehension age equivalent(^b)</td>
<td>21</td>
<td>32</td>
<td>15</td>
<td>17</td>
<td>23</td>
<td>32</td>
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<tr>
<td>Production age equivalent(^b)</td>
<td>14</td>
<td>20</td>
<td>10</td>
<td>17</td>
<td>17</td>
<td>20</td>
</tr>
<tr>
<td>Nonverbal developmental level(^c)</td>
<td>22</td>
<td>25</td>
<td>16</td>
<td>20</td>
<td>24</td>
<td>23</td>
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<tr>
<td>Maternal</td>
<td></td>
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<tr>
<td>Age</td>
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<td>38</td>
<td>36</td>
<td>35</td>
<td>26</td>
<td>27</td>
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<tr>
<td>IQ(^d)</td>
<td>98</td>
<td>113</td>
<td>100</td>
<td>126</td>
<td>108</td>
<td>97</td>
</tr>
</tbody>
</table>

\(^a\)MacArthur-Bates Communicative Development Inventory: Words and Gestures (Fenson et al., 2007). Raw scores from the 396-item vocabulary checklist are reported. \(^b\)Preschool Language Scales–Fourth Edition (Zimmerman, Steiner, & Pond, 2007). Age-equivalent scores are reported for the Auditory Comprehension and Expressive Communication subscales. \(^c\)Mullen Scales of Early Learning (Mullen, 1995). Age-equivalent scores are averaged across the Visual Reception and Fine Motor subscales. \(^d\)Kaufmann Brief Intelligence Test–Second Edition (Kaufman & Kaufman, 2004).
clinician (the second author) who was completing her clinical fellowship year. The clinician also served as the coach for each family. Materials included written handouts and accompanying PowerPoint (Microsoft, Redmond, WA) presentations with embedded video clips. During parent education lessons, parents also watched and discussed video clips of parent–child interaction from previous coaching sessions.

Lesson 1 (Intervention Week 1) introduced the use of verbal descriptions of the child’s focus of attention (i.e., follow-in commenting), use of preferred activities, and noncontingent reinforcement to increase engagement in play routines. For example, if the child were putting blocks into a container, the mother would be encouraged to say something such as “Red block in!” Lesson 2 (Intervention Week 5) introduced the use of indirect strategies to prompt child communication acts (i.e., environmental arrangement, time delay, and choice making). For example, when helping her child with a puzzle, the mother might be encouraged to keep all of the pieces in her lap and offer the child one piece at a time while keeping that piece in sight but out of reach of the child. Lesson 3 (Intervention Week 9) introduced the use of contingent verbal responding to child communication acts (i.e., interpreting nonverbal communication acts and expanding verbal communication acts). For example, if the child reached for the horse puzzle piece, the mother might be encouraged to say “Horse! Neigh!” and then give the piece to the child. Lesson 4 (Intervention Week 13) introduced the use of interactive book reading and open-ended questions to prompt child communication to which the parent could contingently respond by interpreting or expanding. For example, if a specific word was in a child’s repertoire, the parent could ask a wh– question to elicit that word, which would then provide the parent with an opportunity to expand.

Parent education lessons provided opportunities for the clinician to use the following coaching behaviors adapted from Friedman, Woods, and Salisbury (2012): conversation and information sharing; direct teaching of parent knowledge or skills using print, video, or verbal information; demonstration of targeted parent strategies accompanied by explanation; and active listening and joint problem solving and reflection with parent.

Face-to-Face Parent–Child Coaching

A face-to-face clinician coaching session was provided immediately following each onsite parent education session (Intervention Weeks 1, 5, 9, and 13). An additional onsite coaching session, but no parent education, was provided as a booster session at the final onsite visit (Intervention Week 17). During onsite coaching, the clinician was in the room with the dyad and observed the mother, demonstrated strategy use with the child, and focused on helping the mother engage her child in play and recognize opportunities for using the responsive verbal interaction strategies. The clinician provided (a) suggestions for what the mother might do (e.g., “Try handing him one of the blocks”), (b) verbal prompts for targeted strategy use (e.g., “Go ahead and interpret that point”) or “This would be a good time to give him a choice”), (c) verbal models of what the parent might say (e.g., “Do you want cow or bear?”), and (d) reinforcement in the form of descriptive praise for a verbal or physical act by the parent (e.g., “Great expansion of blue!”). Thus, each onsite coaching session represented a series of ongoing opportunities for the mother to practice strategy use with her child while receiving feedback from the clinician.

Distance Parent Coaching

Twelve distance sessions were provided for each dyad, with one distance coaching session held during each week of the month following an onsite visit (Study Weeks 2, 3, 4, 6, 7, 8, 10, 11, 12, 14, 15, and 16). Distance coaching sessions were implemented by the speech-language clinician and provided opportunities for the caregiver to practice targeted strategies while receiving feedback from the clinician as the play occurred in real time in the home.

Fidelity of Clinician Coaching

The licensed SLP who implemented the parent education lessons and the speech-language clinician who implemented the onsite and distance coaching sessions had been trained during a previous parent-mediated intervention project (McDuffie et al., 2013). To evaluate the fidelity of coaching and performance feedback procedures, two trained observers who were not involved in delivering the intervention used a six-item checklist, adapted from Friedman et al. (2012), to code the use of coaching strategies during parent education lessons and onsite and distance coaching sessions. Four randomly selected sessions (one parent education lesson, one onsite coaching session, and two distance coaching sessions) were coded for each dyad using a 30-s partial interval coding procedure. Fidelity coding was completed for 30 min of each randomly selected parent education lesson and 20 min of each randomly selected onsite and distance coaching session. Mean interobserver agreement was above 80% for each item.

Intervention Targets

Follow-In Commenting

This category of verbal responsiveness was defined as parent utterances that followed into the child’s focus of attention and verbally described what the child was doing or playing with immediately prior to the parent response (McDuffie & Yoder, 2010). Follow-in commenting does not convey an expectation that the child provide a behavioral or communicative response to the parent.

Indirect Communication Prompting

Prompting strategies included environmental arrangement (Hancock & Kaiser, 2006), time delay (Warren & Kaiser, 1986), and providing opportunities for choice making within and between activities (McCormick, Jolivette, & Ridgley, 2003). These prompts were considered to be indirect because they did not involve imitation prompts (e.g., “Say _____”) or questions (e.g., “What do you want?”).
Promoted and Spontaneous Child Communication Acts

Promoted child communication acts included any child nonverbal or verbal acts of intentional communication that followed within 3 s of the use of environmental arrangement, choice making, or time delay by the parent. Spontaneous child communication acts included all nonverbal and verbal child acts of intentional communication that were not imitated or promoted by the mother.

Parent Verbal Responses to Child Communication Acts

Parents were taught to respond contingently by interpreting nonverbal communication acts or expanding verbal communication acts. When interpreting, the parent uses a noun, verb, and/or function word to linguistically encode the presumed meaning of the child’s nonverbal communication act (Yoder & Warren, 2001). When expanding, the parent adds semantic or grammatical information in response to a child’s imitative, prompted, or spontaneous verbal utterance (Yoder, Spruytenburg, Edwards, & Davies, 1995).

Data Collection, Coding, and Reliability

Data on maternal strategy use and child communication acts were collected during the 10-min baseline play samples and for 10 min at the approximate midpoint of each face-to-face or distance coaching session. Thus, data were collected during five onsite coaching sessions (Intervention Weeks 1, 5, 9, 13, and 17) and during 12 distance coaching sessions (Intervention Weeks 2, 3, 4, 6, 7, 8, 10, 11, 12, 14, 15, and 16). During data collection, the clinician left the room (onsite) or muted the laptop microphone (distance) and did not interact with or provide feedback to the mother.

For both primary and reliability coding, data were coded from videotape using ProcoderDV (http://procoder.vueinnovations.com), a personal computer–based software program that allows accurate coding of observational data from digital media files (Tapp, 2003). Partial interval coding utilizing a two-step coding process was used to code follow-in commenting. On the first pass through the session video, the coder determined whether the child was engaged or not engaged during each 5-s interval. On the next pass through the video, the coder determined whether or not the mother had used follow-in commenting during each interval that had been judged as engaged. Data for this variable are reported as the percentage of engaged intervals during which the mother used follow-in commenting. The remaining dependent variables were coded as frequency counts. Indirect prompting represents the summed frequency with which parents used environmental arrangement, choice making, and time delay. Prompted child communication acts represent the summed frequency of nonverbal and verbal child communication acts that followed within 3 s of a parent prompt. Spontaneous child communication acts represent the summed frequency of nonpromoted verbal and nonverbal communication acts. Parent contingent verbal responses represent the summed frequency with which mothers used either interpreting or expanding within 3 s of a child communication act.

Dependent variables were coded from digitized videotapes of baseline sessions and from the 10-min portion of each onsite and distance session during which no coaching was provided. The speech-language clinician completed all primary coding, and a second trained observer independently coded 20% of baseline and treatment sessions that were randomly selected. For engagement, an agreement was counted when both coders agreed that the same interval was judged as engaged or not engaged. For follow-in commenting, both coders had to agree that this target behavior happened within the same 5-s interval. For the remaining dependent variables, which were frequency counts, both observers had to independently agree that a given behavior occurred within a 2-s window. Thus, the records for both observers were compared on a point-by-point basis, and percentage agreement was calculated as number of agreements divided by number of agreements plus disagreements. This quotient was then multiplied by 100 and averaged across all dyads. Mean interobserver agreement was 92% for engaged intervals, 90% for follow-in commenting, 87% for indirect communication prompts, 82% for verbal responses to child communication acts, 84% for child-prompted communication acts, 87% for child spontaneous communication acts, and 81% for total child communication acts. The data presented in Table 2 represent interobserver agreement pooled across each phase of the intervention (baseline and treatment) for each dyad.

Results

Intervention Effects on Maternal Verbal Responsivity and Child Communication Acts

Follow-In Commenting

Follow-in commenting was introduced during the first onsite intervention session (Intervention Week 1). Figures 1 and 2 display the frequency of 5-s intervals during which mothers used follow-in commenting (i.e., verbally described their child’s focus of attention) during baseline, intervention, and 3-month follow-up phases for Dyads 1 through 3 and 4 through 6, respectively. During baseline, each mother engaged in relatively low and moderately varied rates of follow-in commenting (follow-in commenting: \( M = 15.73 \), range = 2–32). Mothers in Dyads 1 and 4 demonstrated an increasing trend in use of follow-in commenting immediately prior to introduction of the intervention. The use of follow-in commenting immediately increased in level with the introduction of this strategy during the first onsite intervention session for the mothers in Dyads 1, 3, 4, and 5. For the mother in Dyad 2, use of follow-in commenting showed a large increase in level during the second distance session, whereas the mother in Dyad 6 increased her use of follow-in commenting during the first distance session. With the exception of the mother in Dyad 4, use of follow-in commenting temporarily declined following the immediacy effect and then increased and remained moderately
variable throughout the remainder of the intervention phase. The mother in Dyad 4 used follow-in commenting at relatively high levels throughout the intervention phase, with the exception of Onsite Visit 3, when contingent responding to child communication acts was introduced. Some overlap exists between maternal use of follow-in commenting during the baseline and intervention phases, but an overall mean change in level was observed for all of the mothers (indirect prompting: $M=0.10$, range = $0–5$), and child-prompted communication acts were consistently observed at low to zero levels across the six children (prompted child acts: $M=0.08$, range = $0–4$). Within the first several weeks following the introduction of this strategy during Onsite Visit 2 (Intervention Week 5), each mother’s use of indirect prompting increased above baseline levels. Likewise, a small to large immediacy effect was observed in prompted communication acts for each of the six children. Following the immediacy effect, responding for both mothers and children increased in variability, with moderate to high levels of variability observed across dyads for the remainder of the intervention phase. Overlap exists between baseline and intervention phases, but an overall mean change in level was observed for the mothers (indirect prompting: $M=4.15$, range = $0–13$) and children (prompted child acts: $M=2.86$, range = $0–12$) for each of the dyads. Maternal use of indirect prompting strategies maintained at 3 months following the intervention for each dyad. Prompted child communication was maintained for the children in

### Table 2. Interobserver agreement for each dependent variable.

<table>
<thead>
<tr>
<th>Dyad</th>
<th>Dependent variable</th>
<th>Baseline M %</th>
<th>Range</th>
<th>Intervention M %</th>
<th>Range</th>
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<tr>
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<td>80–90</td>
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<td>88</td>
<td>84–92</td>
</tr>
<tr>
<td></td>
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<td>88–97</td>
<td>92</td>
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<tr>
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<td>84</td>
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</tr>
<tr>
<td></td>
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<td>80–90</td>
</tr>
<tr>
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<td>88–94</td>
</tr>
<tr>
<td></td>
<td>Parent interprets/expands</td>
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<td>92–96</td>
<td>90</td>
<td>88–92</td>
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<tr>
<td></td>
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<td>Parent interprets/expands</td>
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<td>84–90</td>
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<td>89–94</td>
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<td>87</td>
<td>81–90</td>
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<tr>
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<td>Parent interprets/expands</td>
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<td>Child total communication acts</td>
<td>86</td>
<td>80–88</td>
<td>88</td>
<td>84–92</td>
</tr>
</tbody>
</table>

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**Indirect Strategies for Prompting Communication**

Indirect prompting strategies were introduced to the mothers during the second onsite intervention session (Intervention Week 5). Figures 3 and 4 display the frequencies with which (a) mothers used indirect communication prompting strategies and (b) children responded by producing a nonverbal or verbal communication act during baseline, intervention, and 3-month follow-up phases for Dyads 1 through 3 and 4 through 6, respectively. During baseline, maternal use of indirect prompting strategies was consistently very low to nonexistent across the six mothers (indirect prompting: $M=0.10$, range = $0–5$), and child-prompted communication acts were consistently observed at low to zero levels across the six children (prompted child acts: $M=0.08$, range = $0–4$). Within the first several weeks following the introduction of this strategy during Onsite Visit 2 (Intervention Week 5), each mother’s use of indirect prompting increased above baseline levels. Likewise, a small to large immediacy effect was observed in prompted communication acts for each of the six children. Following the immediacy effect, responding for both mothers and children increased in variability, with moderate to high levels of variability observed across dyads for the remainder of the intervention phase. Overlap exists between baseline and intervention phases, but an overall mean change in level was observed for the mothers (indirect prompting: $M=4.15$, range = $0–13$) and children (prompted child acts: $M=2.86$, range = $0–12$) for each of the dyads. Maternal use of indirect prompting strategies maintained at 3 months following the intervention for each dyad. Prompted child communication was maintained for the children in
Dyads 1, 4, and 5; however, prompted child communication acts at the 3-month follow-up were observed at zero or near-zero levels for the children in Dyads 2, 3, and 6. In terms of the size of the intervention effect across dyads, the mean nonoverlap index (NAP) for maternal use of indirect prompting and child-prompted communication acts was 88% and 87%, respectively, both indicating a moderate effect of the intervention. For each dyad, means, ranges, and values of NAP for parent prompts and child-prompted communication acts are individually presented in Table 3.

**Child Spontaneous Communication Acts**

Intervention effects on the spontaneous production of nonverbal and verbal communication acts by child participants were evaluated relative to the first onsite coaching session. Means, ranges, and NAP scores for spontaneous child communication acts are individually presented for each child in Table 3. During baseline, children produced low levels of spontaneous communication acts (child spontaneous communication: \( M = 2.51, \text{ range} = 0-9 \)), with a slight improvement during intervention sessions (\( M = 4.24, \text{ range} = 0-18 \)). However, the child in Dyad 3 had almost no communication acts during baseline and continued to produce very few unprompted communication acts during the intervention, yielding an NAP index of .55. This value suggests almost complete overlap of baseline and intervention data. Two children (Dyads 1 and 6) achieved NAP scores of 64% and 67%, respectively, indicating weak intervention effects. Two children (Dyads 4 and 5) produced very few spontaneous communication acts during baseline but did slightly increase their production of these acts following the introduction of the intervention (Intervention Week 1), resulting in NAP scores of 70% and 79%, respectively, suggesting moderate intervention effects. Last, the child in Dyad 2, who had the most frequent production of spontaneous communication acts during baseline (child spontaneous
communication: \( M = 3.88, \text{ range } = 1–5) \) increased his production of spontaneous communication acts relative to baseline \( (M = 7.64, \text{ range } = 0–23) \), with an NAP index of 72\%, a moderate intervention effect.

**Contingent Verbal Responses to Child Communication Acts**

Contingent responding to child communication acts was introduced to the mothers during the third onsite intervention session (Intervention Week 9). For Dyads 1 through 3 and 4 through 6, Figures 5 and 6, respectively, display frequencies with which children produced communication acts and mothers responded contingently to these acts by either interpreting or expanding. In these figures, the value for child communication acts is a composite metric representing the frequency of both spontaneous and prompted communication acts (either verbal or nonverbal).

During baseline, maternal responses to child communication acts varied across participants \( (M = 2.48, \text{ range } = 0–15) \). With the exception of Dyads 2 and 6, contingent maternal responses during baseline closely mirrored the varied level and trend observed for child communication acts prior to the introduction of this parent language support strategy. The child in Dyad 3 produced almost no communication acts, thus limiting maternal opportunities to respond contingently. Upon introduction of the contingent responding strategy during the third parent education session (Intervention Week 9), decreased child communication and decreased maternal responding were observed for Dyad 1, although an immediacy effect was observed for Dyads 2 and 6, followed by a decelerating trend. Responding during the remaining intervention sessions was moderately to highly variable, with considerable overlap between baseline and intervention periods.
and intervention phases. Child and maternal responding were high at the 3-month follow-up only for Dyads 4, 5, and 6. Overall, visual inspection and consideration of NAP values reflect the lack of a functional relationship between the intervention and contingent maternal responses to child communication acts for Dyads 1 and 5 and a moderate effect of the intervention for Dyads 2, 3, 4, and 6. Table 4 displays individual means, ranges, and values of NAP for each dyad.

### Efficacy of Distance Video-Teleconferencing in Delivering Parent Coaching

Means for onsite coaching sessions were derived by summing the total frequency for each category of parent strategy use and dividing by five (the number of onsite sessions). Means for distance coaching sessions were derived by summing the total frequency for each category of parent strategy use and dividing by 12, with the exception of Dyad 1, who participated in 11 sessions. Nonparametric Wilcoxon signed-ranks tests failed to reveal any significant differences between settings for any of the targeted parent strategies, although strategy use during distance sessions was most consistent for follow-in commenting. Median scores for parent use of follow-in commenting during onsite and distance sessions were 30.30 and 33.96, respectively ($Z = -0.734, \ p = .463$). For parent use of communication prompts, median scores for onsite and distance sessions were 2.80 and 2.42, respectively ($Z = -1.57, \ p = .116$). Median scores for parent verbal responses to child communication acts during onsite and distance sessions were 7.90 and 4.46, respectively ($Z = -1.57, \ p = .116$). These findings suggest that, on average, parent use of strategies introduced during onsite visits was maintained during distance sessions.

Following the intervention, parents were mailed a 15-item survey aimed at assessing the acceptability and feasibility of the parent education lessons as well as face-to-face and distance coaching sessions. Parents were asked to return their anonymous surveys in a postage-paid envelope. For each item in the survey, a 5-point Likert scale provided numerical ratings (1 = strongly disagree, 5 = strongly agree). Higher ratings reflect greater acceptability and feasibility. All six mothers returned their surveys; the overall mean of responses across items and dyads was 4.80/5 (see Table 5).

### Discussion

Delays in spoken language and impairments in the use of language for social communication purposes are a phenotypic characteristic of boys with FXS. In addition,
some mothers of children with FXS may be predisposed to challenges in psychological well-being that may negatively affect verbal responsiveness, especially in the context of raising a child with a disability and high rates of challenging behavior. Thus, developing approaches to optimize patterns of parent–child interaction and support mothers as they scaffold their child’s language development is a high priority for research and practice. The intervention implemented in the current was designed to teach mothers of young boys with FXS to use a set of responsive verbal interaction strategies to increase the amount of appropriately scaffolded verbal language input that each child received. In general, the findings suggest that despite the challenges faced by these dyads, the intervention led to meaningful and positive changes in both maternal and child targeted behaviors.

All of the maternal strategies targeted in the current study were selected for their utility in providing increased levels of salient and easily processed verbal language to children. In follow-in commenting, the mother was encouraged to provide verbal language input that directly corresponded to the child’s ongoing focus of attention (McDuffie & Yoder, 2010). We found a moderately beneficial effect of the intervention on maternal use of follow-in commenting. In fact, all mothers showed increases over baseline, although at different points during the intervention. Follow-in commenting establishes a state of symbol-infused supported joint engagement in which the child receives valuable contingent linguistic information while not being required to contribute actively to the maintenance of this triadic interaction state (Adamson, Bakeman, & Deckner, 2004; Adamson, Bakeman, Deckner, & Romski, 2009). Parent
use of follow-in commenting has been shown to predict gains in both comprehension and spoken language outcomes for developmentally young and minimally verbal children with ASD (Haebig et al., 2013). Thus, the efficacy of the present intervention in increasing maternal follow-in commenting has the potential to lead to meaningful improvement in language outcomes for developmentally young children with FXS. That two of the mothers in our sample did not maintain their use of this strategy after the end of coaching, however, suggests that some mothers may need more instruction or practice in this strategy or more options regarding situations in which to use it.

Mothers in the current study were also taught strategies for indirectly prompting child communication acts through the use of environmental arrangement, time delay, and choice making. Such indirect prompts capitalize on child motivation to increase requests for the desired object or to continue a highly preferred activity or routine. We found that the intervention led to moderate increases in maternal use of indirect prompts over baseline, with all mothers showing and sustaining this improvement at intervention follow-up. Indirect prompting is likely to be especially important for children with FXS because many are infrequent communicators (Abbeduto et al., 2007), which means that they provide few opportunities for their parents to respond with the types of contingent interpretations or expansion that help advance language learning.

In conjunction with increased maternal prompting, all of the child participants also showed an increased frequency of prompted communication acts over baseline. This is an important finding because, when responding contingently to these acts, parents have more opportunities to provide the rich linguistic input from which the children can abstract new vocabulary and combinatorial rules. At
the same time, however, we found that children did not respond to all—or even most—indirect prompts from their mothers, and half of the children failed to maintain their gains at follow-up. In the intervention, mothers were taught to use prompting as a way of continuing the child’s ongoing play, but continuation of play might not provide sufficient reinforcement for the child with FXS. Parents might benefit from coaching focused on successfully contriving transitive conditioned motivating operations that will temporarily increase the value of the reinforcer and the likelihood of child communication (Carbone, 2013). Children with FXS also may require more direct physical or echoic prompts to ensure communication following an indirect prompt, with gradual fading of the prompts via time delay (Carbone, Sweeney-Kerwin, Attanasio, & Kasper, 2010). In addition, children with FXS who present with limited functional communication may benefit from mand training with manual signs that can be reliably prompted. Research with children with ASD suggests improved speech production and comprehension as a result of using this approach (e.g., Carbone et al., 2010; Schlosser & Wendt, 2008; van der Meer, Sutherland, O’Reilly, Lancioni, & Sigafoos, 2012). Thus, future research could focus on the potential benefit of a presession warm-up activity consisting of structured mand training with high-magnitude reinforcement (e.g., food or access to highly preferred toy) followed by the naturalistic intervention sessions.

The intervention had modest and variable effects on the children’s spontaneous (unprompted) communication acts. Five of the children showed increased frequency of unprompted communication acts relative to baseline during the intervention. For three of these children the effects were moderate, but for two the effects were minimal. One child showed no meaningful change in unprompted communication as a result of the intervention. As indicated...
previously, it may be necessary to provide more direct scaffolding on very direct maternal prompts for communication to achieve larger increases in child (prompted and unprompted) communication acts. Nonetheless, it is notable that five of the six children were induced to be more spontaneously communicative by this relatively brief intervention. Increased child communication means that there will be more opportunities for parents to engage in language “teaching” via contingent responding. Moreover, it may be that even a rather minimal improvement in communicativeness can have a powerful effect on parental perceptions of the child and on the parent’s feelings of her own parenting competence.

The children in the intervention who produced minimal communication acts (either spontaneous or prompted) may have benefited from an augmentative communication approach. Although the use of manual signs and communicative gestures was encouraged throughout the intervention, none of the children had access to an aided augmentative and alternative communication device through this intervention program, which may have served to increase both prompted and spontaneous communication acts. Last, stimulus–stimulus pairing (Esch, Carr, & Grow, 2009; Rader et al., 2014), a procedure in which a clinician’s targeted vocalizations are produced in child-directed language and paired with the delivery of highly preferred items, may have been useful in increasing the frequency of spontaneous vocalizations or verbalizations during pretreatment sessions. Through the use of the stimulus–stimulus pairing procedure, child vocalizations acquire reinforcing properties due to their association with the preferred item and become automatic reinforcers to the child who produces them. Vocal responses acquired in this way might then be reinforced during subsequent intervention sessions.
The final targeted parent language-facilitation strategy was the provision of verbal language input contingently in response to child communication acts. Thus, mothers were taught to interpret nonverbal communication acts and to expand verbal communication acts. Four of the six mothers did increase the frequency with which they responded. However, the number of opportunities for mothers to use this strategy was limited by the number of spontaneous and prompted child communication acts that were produced. We have already mentioned that more direct prompting of child communication may have been necessary to support child responding. Whether this level

<table>
<thead>
<tr>
<th>Dyad</th>
<th>Child total communication acts*</th>
<th>Parent contingent verbal responses*</th>
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<td></td>
<td>NAP</td>
<td>.74</td>
</tr>
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</table>

Note. NAP = nonoverlap of all pairs.

*aNine intervention sessions.

The final targeted parent language-facilitation strategy was the provision of verbal language input contingently in response to child communication acts. Thus, mothers were taught to interpret nonverbal communication acts and to expand verbal communication acts. Four of the six mothers did increase the frequency with which they responded. However, the number of opportunities for mothers to use this strategy was limited by the number of spontaneous and prompted child communication acts that were produced. We have already mentioned that more direct prompting of child communication may have been necessary to support child responding. Whether this level

Table 5. Results of the parent satisfaction survey.

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<th>Overall program</th>
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<td>5</td>
</tr>
<tr>
<td>Recommend to other parents of children with FXS</td>
<td>5.00</td>
<td>5</td>
</tr>
<tr>
<td>I have learned strategies that are useful when I interact with my child.</td>
<td>4.83</td>
<td>4–5</td>
</tr>
<tr>
<td>I am able to engage my child in play more easily than prior to the intervention.</td>
<td>4.83</td>
<td>4–5</td>
</tr>
<tr>
<td>My child is able to communicate more successfully than prior to the intervention.</td>
<td>4.83</td>
<td>4–5</td>
</tr>
<tr>
<td>My child’s attention span is longer than it was prior to the intervention.</td>
<td>4.67</td>
<td>4–5</td>
</tr>
<tr>
<td>I learned information that was relevant to my child’s communication ability.</td>
<td>5.00</td>
<td>5</td>
</tr>
<tr>
<td>I learned information that was relevant to my child’s behavioral challenges.</td>
<td>5.00</td>
<td>5</td>
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<tr>
<td>Overall, I am satisfied with my experiences in this program.</td>
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<tr>
<td>The PowerPoint presentations were useful.</td>
<td>4.33</td>
<td>3–5</td>
</tr>
<tr>
<td>Overall, I was satisfied with the parent education sessions.</td>
<td>4.83</td>
<td>4–5</td>
</tr>
<tr>
<td>The coaching sessions enabled me to practice the targeted strategies.</td>
<td>4.67</td>
<td>4–5</td>
</tr>
<tr>
<td>Overall, I was satisfied with the onsite coaching sessions.</td>
<td>4.50</td>
<td>4–5</td>
</tr>
<tr>
<td>I thought the distance coaching sessions were helpful.</td>
<td>4.67</td>
<td>4–5</td>
</tr>
<tr>
<td>It was easy to use the computer for distance coaching sessions.</td>
<td>5.00</td>
<td>5</td>
</tr>
</tbody>
</table>

Note. 1 = strongly disagree, 3 = neither agree nor disagree, 5 = strongly agree.
of parental interpretation and expansion is sufficient to boost language learning in severely impaired children such as those studied here remains to be determined.

Maternal use of follow-up prompting of communication showed more robust improvement relative to changes in responding contingently to child communication acts. There are two potential explanations for this finding. As mentioned previously, children whose initial levels of communication were very low showed variable levels of prompt responding and did not show improvements in the production of spontaneous communication acts. Thus, some mothers simply did not have many opportunities to respond contingently to child communication acts relative to baseline. In addition, examination of the results for contingent responding to child communication acts (see Figures 5 and 6) suggests that mothers began to increase contingent responding to child communication acts as soon as they were taught prompting strategies in Parent Education Session 2 (Intervention Week 5). Thus, it does not appear that there was functional independence between parent prompting and parent contingent responses to child communication acts. It is possible that, in the process of learning follow-up commenting and indirect strategies to prompt communication, mothers became more responsive in general and began to respond to child communication acts. It also is possible that when mothers learned to use indirect prompting strategies, they also became more aware of or able to identify communication acts that their children produced. At least four of the dyads showed an abrupt increase in maternal contingent responding during the last four sessions prior to the introduction of the contingent responding strategy. This time period represents the onsite and distance sessions during which prompting strategies were introduced and targeted. In the current study, the decision was made to introduce the maternal strategies one at a time to not overwhelm the mothers. In future studies, it might be preferable to introduce all three targeted strategies at one time to avoid the lack of independence between the dependent variables that arose in the current study.

The fact that maternal use of targeted intervention strategies was comparable during both onsite and distance sessions lends preliminary support to the premise that parent coaching sessions can be implemented effectively by means of distance video-teleconferencing. We had demonstrated previously the effectiveness of distance delivery of a parent–implemented language intervention for mothers of children with ASD (McDuffie et al., 2013). Although not significantly different between settings, the data suggest that parent strategy use was somewhat more consistent during onsite relative to distance sessions. Our clinical impression is that the clinic setting may have been less distracting for the children; that is, it may have been easier to keep children productively engaged with both the task and the caregiver at the clinic because the testing room was small and contained only a few highly interesting toys. In addition, it is possible that mothers were more “on point” during onsite visits than they were at home, where they may have been preoccupied with daily obligations. Overall, however, these results do suggest that distance technology holds great promise for the delivery of early language interventions and that more research is needed to refine the use of this tool and take advantage of its potential.

Limitations of the Current Study

The current study had several limitations. Because of the time constraints of the onsite preintervention visits, which lasted only 2 days, several baseline sessions were collected on the same day for each participant. Thus, the baseline data may not have been representative of child language performance over a more extended period of time. However, it was our clinical impression that baseline sessions did accurately reflect the children’s level of communication development. As a pilot study, the intervention included only six mother–child dyads and provided only 16 weekly sessions over 4 months. Although mothers did learn to use new skills, the brief time frame of the intervention may have constrained the ability of the mothers to practice and consolidate newly learned skills and precluded the likelihood of observing more distal effects on child communication. Although the desirability of using targeted strategies in contexts other than play was discussed frequently with mothers, we did not directly sample the use of targeted strategies in daily routines. A further limitation was that a predetermined number of coaching sessions was provided between each parent education lesson. It is likely that different mothers needed different lengths of time to become proficient in using targeted strategies and that a criterion level of performance benchmarks should have been used (e.g., M. Roberts & Kaiser, 2012) prior to the introduction of new strategies. Given the short time frame of the intervention, we do not present standardized test scores at postintervention because these global measures are unlikely to reflect changes in child communication that are attributable to the intervention. Last, although the current study enrolled only mothers, it is important to emphasize that an intervention that included fathers might further enrich the child’s language learning environment and might provide mothers with social support in addressing the developmental needs of their children.

Future Directions

Several child characteristics substantially influenced maternal ability to implement the targeted intervention strategies successfully. Chief among these child factors were challenging behaviors, observed both clinically and empirically (e.g., tantrums, throwing, hitting, and self-injury; Machalicek et al., 2014), that negatively affected the amount of time children were actively engaged during coaching sessions. Opportunities for mothers to use targeted strategies also were limited by child inattention and limited interest in toys. A child’s willingness to engage with objects theoretically can provide a conceptual platform onto which verbally responsive caregiver language can be mapped in
support of language development (Bottema-Beutel et al., 2014). Thus, parents will have fewer opportunities to provide follow-in language to the extent that children are not interested in, or motivated to engage with, objects. In addition, it was found in a previous study that the children enrolled in the current study all demonstrated escape and tangibly maintained challenging behaviors (e.g., hitting, crying, elopement; Machalicek et al., 2014) and limited object play skills (McDuffie et al., 2015). Future iterations of the intervention should focus on decreasing challenging behaviors and maximizing child engagement as a component of the parent-mediated intervention.

It will be important for researchers to examine whether a greater proportion of intervention sessions could be provided at a distance and to determine how best to support each mother as she implements the language-facilitation strategies in the home. In the current study, families were asked to travel substantial distances to attend a monthly onsite visit at the research center. Although none of the mothers in this study missed an onsite visit, it would clearly be more feasible in terms of effort and cost if the majority of the intervention could be provided at a distance rather than through a combination of distance and onsite sessions. Improvements in access to broadband Internet and in the quality of teleconferencing software should make an intervention delivered completely at a distance more attainable in the near future. Last, the number, duration, and weekly distribution of distance sessions needed to achieve optimal outcomes and high levels of acceptability to families remains to be determined.

Many families with FXS have more than one affected child. The current study was designed to provide the intervention to each mother during interaction with only one of her children. An important focus of future research will be to refine procedures for delivering the intervention as the mother interacts with more than one child because this approach would be of greater utility to families of children with FXS. Future research efforts should be directed toward establishing and evaluating methods for providing distance coaching during family routines other than play. The importance of providing children with responsive verbal language input during daily routines cannot be overstated and deserves continued attention as we develop and refine procedures for the use of distance teleconferencing. Last, clinical trials are now underway to identify pharmacological treatments that will remediate the core deficits of individuals with FXS (Berry-Kravis, Knox, & Hervey, 2011). An important question to consider is whether the effects of a pharmacological intervention can be moderated by the addition of a behavioral intervention.

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