

# UC Davis

## UC Davis Previously Published Works

### Title

A Spoken-Language Intervention for School-Aged Boys With Fragile X Syndrome

### Permalink

<https://escholarship.org/uc/item/33917618>

### Journal

American Journal on Intellectual and Developmental Disabilities, 121(3)

### ISSN

1944-7515

### Authors

McDuffie, Andrea  
Machalicek, Wendy  
Bullard, Lauren  
[et al.](#)

### Publication Date

2016-05-01

### DOI

10.1352/1944-7558-121.3.236

Peer reviewed



Published in final edited form as:

*Am J Intellect Dev Disabil.* 2016 May ; 121(3): 236–265. doi:10.1352/1944-7558-121.3.236.

## A Spoken Language Intervention for School-Aged Boys with fragile X Syndrome

Andrea McDuffie<sup>1</sup>, Wendy Machalicek<sup>2</sup>, Lauren Bullard<sup>1</sup>, Sarah Nelson<sup>1</sup>, Melissa Mello<sup>1</sup>, Robyn Tempero-Feigles<sup>1</sup>, Nancy Castignetti<sup>1</sup>, and Leonard Abbeduto<sup>1</sup>

<sup>1</sup>Department of Psychiatry and Behavioral Sciences and UC Davis MIND Institute

<sup>2</sup>University of Oregon

### Abstract

Using a single case design, a parent-mediated spoken language intervention was delivered to three mothers and their school-aged sons with fragile X syndrome, the leading inherited cause of intellectual disability. The intervention was embedded in the context of shared story-telling using wordless picture books and targeted three empirically-derived language support strategies. All sessions were implemented via distance video-teleconferencing. Parent education sessions were followed by 12 weekly clinician coaching and feedback sessions. Data was collected weekly during independent homework and clinician observation sessions. Relative to baseline, mothers increased their use of targeted strategies and dyads increased the frequency and duration of story-related talking. Generalized effects of the intervention on lexical diversity and grammatical complexity were observed. Implications for practice are discussed.

### Keywords

fragile X syndrome; spoken language intervention; shared book reading; parents

---

Boys with fragile X syndrome (FXS), the leading inherited cause of intellectual disability, display marked delays in the ability to use spoken language for effective interpersonal communication. Delays in spoken communication, which continue into adolescence and adulthood (Abbeduto, Brady, & Kover, 2007), negatively impact the ability to function independently in academic, social, and vocational settings (Hartley, Seltzer, Raspa, Olmsted,

---

Corresponding Author: Andrea McDuffie, PhD, CCC-SLP, [asmcduffie@ucdavis.edu](mailto:asmcduffie@ucdavis.edu), University of California Davis, Department of Psychiatry and Behavioral Sciences, MIND Institute, 2825 50th Street, Sacramento, CA, USA 95817.

Wendy Machalicek, PhD, BCBA-D, [wmachali@uoregon.edu](mailto:wmachali@uoregon.edu), University of Oregon, Department of Special Education and Clinical Sciences, 352 Clinical Service Building, Eugene, OR, USA 97403

Lauren Bullard, BA, [bullard@ucdavis.edu](mailto:bullard@ucdavis.edu), University of California Davis, Department of Human Development, MIND Institute, 2825 50th Street, Sacramento, CA, USA 95817

Sarah Nelson, MS, [snnelson@ucdavis.edu](mailto:snnelson@ucdavis.edu), University of California Davis, Department of Human Development, MIND Institute, 2825 50th Street, Sacramento, CA, USA 95817

Melissa Mello, MS, BCBA, [bullard@ucdavis.edu](mailto:bullard@ucdavis.edu), University of California Davis, Department of Human Development, MIND Institute, 2825 50th Street, Sacramento, CA, USA 95817

Robyn Tempero-Feigles, BS, [rjtempero@ucdavis.edu](mailto:rjtempero@ucdavis.edu), University of California Davis, Department of Human Development, MIND Institute, 2825 50th Street, Sacramento, CA, USA 95817

Nancy Castignetti, MS, [nacastignetti@ucdavis.edu](mailto:nacastignetti@ucdavis.edu), University of California Davis, Department of Human Development, MIND Institute, 2825 50th Street, Sacramento, CA, USA 95817

Leonard Abbeduto, PhD, [ljabbeduto@ucdavis.edu](mailto:ljabbeduto@ucdavis.edu), University of California Davis, Department of Human Development, MIND Institute, 2825 50th Street, Sacramento, CA, USA 95817

Bishop, & Bailey, 2011). Despite the pressing need for spoken language interventions targeting middle and high school-aged individuals, very few published studies have examined the efficacy or effectiveness of any type of language intervention for adolescents with neurodevelopmental disorders (Cirrin & Gillam, 2008).

In the current pilot study, we used a single-case experimental design to examine the initial promise of a naturalistic, parent-implemented, language intervention designed to improve the spoken language skills of school-aged boys with FXS. We delivered the intervention by means of distance video-teleconferencing to enable families to access the intervention at times that were convenient for their schedule and regardless of geographic location. We taught mothers to support their sons' spoken language as they jointly participated in a back and forth conversation focused on the shared topic of a wordless picture book. As a result of the intervention, we expected to observe positive changes in maternal use of targeted language support strategies as well as gains in children's use of more diverse vocabulary and developmentally advanced grammar.

## The Behavioral Phenotype of fragile X Syndrome

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 transcriptional silencing of the gene, reducing or completely eliminating its protein product, FMRP (Oostra & Willemsen, 2009). FMRP is critical for the maturation and pruning of synapses (Bhakar, Dölen, & Bear, 2012). Males with the *FMR1* full mutation tend to be more affected than females, given the protective presence of an unaffected X chromosome in females.

Virtually all males with the *FMR1* full mutation have cognitive delays, with the vast majority having IQs between 40 and 55 (Hessl et al., 2009). These males+ also demonstrate significant delays in language development (Roberts, Mirrett, Anderson, Burchinal, & Neebe, 2002), even relative to what would be expected based on nonverbal cognitive developmental level (Abbeduto et al., 2007). The majority of males with FXS display hyperactivity and attentional difficulties (Scerif, Longhi, Cole, Karmiloff-Smith, & Cornish, 2012), anxiety and social withdrawal (Cordeiro, Ballinger, Hagerman, & Hessl, 2011), as well as challenging behaviors, including aggression and self-injury (Leigh, Hagerman, & Hessl, 2012; Symons, Clark, Hatton, Skinner, & Bailey, 2003). Most males with FXS also demonstrate symptoms of autism (Hall et al., 2010; Merenstein et al., 1996), with as many as 60% displaying behaviors that are frequent and severe enough to warrant a comorbid diagnosis of an autism spectrum disorder (ASD) (Bailey, Mesibov, Hatton, Clark, Roberts, Mayhew, 1998; Hall, Lightbody, Hirt, Rezvani, & Reiss, 2010).

## Language development in FXS

Language comprehension is delayed in FXS relative to expectations for both chronological age and nonverbal cognitive level (Roberts, Mirrett, & Burchinal, 2001; Price, Roberts, Vandergrift, & Martin, 2007). Similarly, studies using standardized assessments of expressive language also have revealed significant delays and slower rates of growth for

boys with FXS relative to younger TD children matched on nonverbal cognitive level (Estigarribia, Martin, & Roberts, 2013; Martin, Losh, Estigarribia, Sideris, & Roberts, 2013). However, the spoken language challenges experienced by males with FXS extend beyond what is captured by standardized assessments and include the ways in which spoken language is used to communicate during back-and-forth interactions with conversational partners. To provide a more complete picture of the ways in which boys with FXS actually use their spoken language skills in naturalistic interactions, researchers have turned to expressive language sampling.

For a large group of boys with FXS who averaged between 8- and 9-years of age, Price, Roberts, Hennon, Berni, Anderson, & Sideris (2008) derived measures of syntactic complexity and morphological development based on language samples collected during administration of the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DiLavore, & Risi, 2002). After controlling for nonverbal mental age and maternal education, boys with FXS had significantly shorter mean length of utterance and significantly lower scores for grammatical complexity than did younger typically developing children. Additionally, boys with FXS used significantly fewer noun and verb phrases, as well as fewer different types of sentence constructions (e.g., SVO sentences, relative clauses, conjunctions, etc.) than TD comparison boys. Similarly, Estigarribia and colleagues (2011) found that boys with FXS, with and without comorbid ASD and ranging in age from 6- to 16-years, produced fewer morphological endings on verbs (e.g., -ed, -s, copula BE, auxiliaries BE, DO, HAVE) and nouns (e.g., articles a, an, the, plural -s, possessive 's) during a language sample derived from the ADOS than did younger typically-developing children. Between-group differences were significant even after controlling for nonverbal mental age, maternal education, and speech intelligibility (Estigarribia, Roberts, Sideris, & Price, 2011).

Abbeduto and colleagues have developed a standardized approach to evaluating spoken language using expressive language sampling in two different contexts: narration of a wordless picture book and an interview-style conversational interaction with an examiner. In contrast to norm-referenced standardized tests of spoken language, a language sample can yield information about how individuals use spoken language to communicate in interactive situations that require more than pointing to or labeling a picture, or formulating a single sentence. In one study, Kover, McDuffie, Abbeduto, and Brown (2012) found that, although 10- to 17-year old boys with FXS produced longer sentences in narration than in conversation, they produced significantly shorter sentences and used fewer different vocabulary words in the narrative context than did younger typically developing participants, even after controlling for nonverbal mental age. In the narrative story telling context, participants with FXS were also significantly less fluent (i.e., produced a higher proportion mazes, dysfluencies, and repetitions) than younger children with typical development. Thus, studies using expressive language sampling can reveal a more nuanced description of the ways in which boys with FXS use spoken language to communicate in every day settings.

Perseveration is a well-studied challenge to successful spoken communication for males with FXS (Abbeduto, Brady, & Kover, 2007), who are likely to repeat words, phrases, and topics when they speak (Belser & Sudhalter, 2001). The amount of perseverative speech

produced by males with FXS is higher than what is observed for typically developing children matched on language level (Levy et al., 2006) as well as relative to children with other neurodevelopmental disorders (Moss, Oliver, Arron, Burbidge, & Berg, 2009). Perseverative speech is thought to be a defining feature of the spoken language phenotype of males with FXS (Abbeduto & Hagerman, 1997). The presence of perseverative speech has been hypothesized to be related to other aspects of the FXS behavioral phenotype including hyperarousal and attentional impairments which may be exacerbated in situations, such as conversation, that are perceived as socially demanding (Abbeduto et al., 2007). In short, the challenges to effective spoken communication experienced by males with FXS will be especially evident in contexts requiring demanding interpersonal interaction. Thus, situating a language intervention within the context of an ongoing conversational interaction may provide many opportunities to target and ameliorate the challenges to spoken language experienced by adolescent males with FXS.

### Spoken Language Intervention Approach

From early in development, language is learned within the context of back-and-forth interactions focused on topics that are shared between social partners (Bruner, 1985; Sameroff & Fiese, 2000). Additionally, high levels of affectively positive and contingent verbal language input from caregivers can optimize language outcomes for children with neurodevelopmental disorders (Hauser-Cram, Warfield, Shonkoff, & Krauss, 2001), including FXS (Warren, Brady, Sterling, Fleming, & Marquis, 2010; Brady, Warren, Fleming, Keller, & Sterling, 2014). In our clinical experience, mothers of children with FXS, almost without exception, report that it is extremely difficult to engage their verbal sons with FXS in sustained conversational interactions on a shared topic (e.g., describing what happened at school; discussing an upcoming trip). This observation is not surprising given that the behavioral characteristics of boys with FXS (i.e., hyperarousal, inattention, tangential and repetitive speech). However, it is likely that such limitations combined with less than optimal interactions, would have a cumulative negative impact on spoken language growth across childhood and adolescence. In designing the present spoken language intervention, therefore, our goal was to optimize language learning opportunities for boys with FXS by providing a supportive context within which mothers could engage in sustained verbal turn-taking related to a shared topic. To accomplish this goal, we embedded the intervention into the context of shared, or interactive, book reading.

Shared book reading provides a familiar setting for parent/child interactions beginning in early childhood (Snow, Perlmann, & Nathan, 1987). Even as children learn to read, shared book reading activities continue to provide a vehicle for supporting children's oral language skills, language comprehension, and literacy development (Dickinson & Porche, 2011; Green & Klecan Akar, 2012). During shared book reading, the adult verbally relates the story to the child, guides discussions about the story content and, by adding information, asking questions, and prompting child responses, encourages the child to gradually assume more responsibility for telling the story independently (Whitehurst et al., 1994; Van Kleeck, Vander Woude, & Hammett, 2006). For speech/language pathologists, shared book reading represents a dynamic and naturalistic context for delivering a spoken language intervention (Kaderavek & Justice, 2002; Swanson, Fey, Mills & Hood, 2005).

Shared book-reading activities maximize the kinds of experiences that support oral language development in at least three ways: (a) by offering children numerous opportunities to hear diverse vocabulary embedded in a variety of grammatical constructions; (b) by providing visual supports for maintaining shared interest and attention; and (c) by providing numerous opportunities for a sustained conversation around a shared topic (Dickinson et al., 2014). For children with language delays, interventions based on shared book reading result in improvements in structural aspects of spoken language (i.e., knowledge of vocabulary, morphosyntax, and grammar) (Bradshaw, Hoffman, & Norris, 1998; Dale, Crain-Thoreson, Notari-Syverson, & Cole 1996; Yoder, Spruytenburg, Edwards, & Davies, 1995), as well as gains in language comprehension (Dunst, Williams, Trivette, Simkus, & Hamby, 2012; Hogan, Bridges, Justice, & Cain, 2011).

Although we use the term “reading” to describe the process of interacting around the shared topic of an illustrated book, decoding of written text was not a focus of the present study. In fact, we removed the text from the books used during the intervention to shift the emphasis of the parent-child interaction exclusively to the use of spoken language. Our goal was to support a back-and-forth conversation between parent and child in which the child would gradually, through the parent use of targeted scaffolding strategies, assume a more active role in retelling the story.

## Selection of Intervention Strategies

Interactive book reading has a history of use as the context for spoken language interventions for children who are at risk for or experiencing language delays (for review, see Petersen, 2011). However, it is not merely the context of shared book reading, but the ways in which caregivers and teachers support their child’s active participation in this activity that determines whether exposure to shared reading opportunities has a positive impact on children’s language skills (Pentimonti et al., 2012; Zevenbergen & Whitehurst, 2003). In the current study, we targeted three strategies that we believed could be learned by mothers within the context of a parent-implemented intervention delivered by distance teleconferencing and that we anticipated would support spoken language development in adolescent boys with FXS.

### Recasting

Recasting is a contingent conversational response that relates to the central meaning of the immediately preceding child utterance while providing additional or corrective semantic or grammatical content (Fey, 1986). Theoretically, the temporal proximity and semantic overlap between the recast and the child’s original utterance allows the child to implicitly notice the differences between the recast and his own utterance (Warren & Yoder, 1997). If the child notices this difference repeatedly and if he/she is developmentally ready to learn the new linguistic form, the child should reorganize his/her own language system to acquire the new vocabulary word or grammatical structure (Camarata, 1995). Several studies have used recasting during spoken language interventions based on shared book reading (Fey, et al., 1994; Swanson et al., 2005; Tyler & Sandoval, 1994; Yoder et al., 1995; Zevenbergen, Whitehurst, & Zevenbergen, 2003).

## WH-Questions

It is likely that children with intellectual impairments will have problems with story comprehension. One line of research suggests that comprehension is increased and children subsequently produce more sophisticated story retells after answering specific questions about the story (Pratt, Kerig, Cowan, & Cowan, 1988); that is, adult wh-questions can prompt the child to more fully understand and precisely express what is happening in a story (Pentimonti et al., 2012). Additionally, children's recall of verbal material is enhanced when they answer questions that, presumably, focus their attention on relevant aspects of a story and when their mistakes in comprehension are corrected by their conversational partner (Albanese & Antoniotti, 1997). Adult wh-questions are useful in eliciting child verbal utterances that maintain a shared topic of conversation (Yoder, Davies, Bishop, & Munson, 1994). Following the child's response to a wh-question, the parent can continue the conversational turn-taking by providing a semantic or grammatical recast (i.e., vertical structuring; Schwartz, Chapman, Terrell, Prelock, & Rowan, 1985). Answering *why*, *how*, and *what happens next* questions is considered a primary technique for eliciting inferential language from children during story-telling; that is, answering these types of wh-questions can encourage children to compare, contrast, evaluate, and hypothesize about story events beyond what is pictured in the illustrations of a book (Pentimonti et al., 2012).

## Intonation Prompts

An intonation prompt (i.e., cloze procedure or time delay; Bradshaw et al., 1998; Charlop, Schreibman, Thibodeau, 1985) is a language elicitation strategy in which the adult provides the beginning segment of an utterance followed by an expectant pause designed to prompt the child to respond by "filling in the blank" or completing the utterance. Use of an intonation prompt narrows the options for what the child can say and allows the child to take a turn in a conversation by completing the production of an utterance that might be too difficult to produce independently (Bradshaw et al., 1998). The child's response to the use of an intonation prompt is likely to be one word or a short phrase. As with child responses to wh-questions, the parent then has the opportunity to provide a contingent recast by expanding the response elicited by the intonation prompt. Finally, if a child answers a question incorrectly, the parent can provide corrective feedback in the form of an intonation prompt, thus eliciting a child response that is more appropriate in the context of the story. Intonation prompts are frequently incorporated into naturalistic language interventions (Kaiser & Roberts, 2013) and have been targeted in parent-implemented language interventions based on shared book reading (Bellon-Harn, Hoffman, & Harn, 2004; Bradshaw et al., 1998).

## Research Questions

The boys with FXS who participated in the current study could be considered verbally fluent in that they produced 3- to 6-word phrases and sentences to communicate their needs and wants on a daily basis. In general, however, their oral language skills were not sufficient to allow them to participate in extended conversational interactions, such as conveying a personal experience or retelling a coherent story. Based upon the cognitive and language characteristics of males with FXS in this age range, we reasoned that a shared book reading

intervention would provide a developmentally appropriate and ecologically valid context within which caregivers could provide verbally responsive language input to their school-aged children. We expected that the quality and quantity of this type of verbal language input would be more optimal than that which would typically be available to boys with FXS during the daily course of incidental and unstructured spoken interactions with caregivers (Petersen, Gillam, Spencer, & Gillam, 2010). Additionally, we hypothesized that the visual supports provided by the illustrations in each book would encourage child engagement, foster comprehension of the shared topic of conversation, and scaffold participant responses in a way that could not be accomplished during a conversational interaction without visual supports (Miles, Chapman, & Sindberg, 2006). The parent strategies targeted during the intervention were utilized to elicit child verbal responses (i.e., asking questions, Intonation Prompts) or to respond contingently to child utterances by providing more diverse vocabulary and developmentally advanced grammar (i.e., recasts). The study addressed the following research questions:

1. Does participation in the intervention increase the frequency with which parents and children produce story-related utterances?
2. Does participation in the intervention increase parent use of targeted intervention strategies?
3. Does participation in the intervention increase children's use of more diverse vocabulary and more complex grammatical constructions?

## Method

### Participants and Setting

Three boys and their biological mothers participated in the study. Dyads were recruited through postings on a listserv for families affected by FXS. Children met the following criteria: (a) confirmed diagnosis of full mutation FXS; (b) between 10 and 16 years of age; (c) daily use of at least 3-word utterances according to maternal report; (d) English was the primary language spoken in the home; and (e) no uncorrected sensory or motor impairments severe enough to preclude processing and responding to verbal language input. A battery of standardized tests, informant report, and expressive language sampling measures were administered during pre- and post-intervention visits to a university-based research center. Informed consent was obtained and all intervention procedures were approved by the university's Institutional Review Board. Characteristics of each dyad at the pre-intervention visit are presented in Table 1.

### Video-teleconferencing Equipment

Equipment loaned to each family consisted of an 11" MacBook Air™ laptop computer, a 16GB 9.7" iPad Air, and a Plantronics M165 Marque 2 Ultralight Bluetooth Headset. The headset was used during coaching sessions so that the child would not hear the clinician's comments to the mother. The books selected by each mother were digitized, the text removed, and loaded onto the iPad using the Apple iBooks application. The clinician used a 13.3 in MacBook Pro™ laptop computer and initiated video calls using the built-in iSight™ web camera and Skype™ software. Coaching and data collection sessions were captured



using eCamm™ Call Recording Software. A wireless broadband internet connection was used and secured through 128-bit advanced encryption. Homework sessions were independently recorded by the mother using the iSight™ camera and Photobooth™ software application on the MacBook and uploaded to the clinical team using Dropbox™, a cloud-based storage system.

### **Narrative Language Sampling**

Samples of narrative language were collected at pre- and post-intervention visits and served as a measure of generalized treatment effects. For each child participant, a narrative language sample was collected with an unfamiliar examiner as well as with the mother. The narrative language sampling procedures have been developed and tested in previous studies (e.g., Kover et al., 2012).

**Narrative language sample with examiner**—The examiner asked the participant to look at a wordless picture book to get the gist of the story. The participant then told the story page-by-page. The examiner followed a script that maximizes the participant's contribution and avoids constraining the participant's talk. Two books by Mercer Mayer were used: *Frog Goes to Dinner* and *Frog on his Own*. Books were counterbalanced, and each participant received a different book at pre- and post-intervention. This language sampling session represented a generalization context as it involved a different setting, materials, and person than did the intervention sessions. Administration: Untimed, but typically 10 to 15 min.

**Narrative language sample with mother**—The mother was instructed to look through the book with her child and encourage her child to tell the story. Two books by Mercer Mayer were used: *Frog, Where Are You* and *One Frog Too Many*. Books were counterbalanced, and each participant received a different book at pre- and post-intervention. Each mother was kept blind to the narrative procedure with the examiner. This language sampling session represented a generalization context as it involved a different setting and materials than did the intervention sessions. Administration: Untimed, but typically 10 to 15 min.

### **Design of the Intervention Program**

The intervention used a single-subject multiple probe design across participants (Gast, Lloyd, & Ledford, 2014). In this design, probe data is collected intermittently during baseline in place of the continuous measurement of baseline data. The design requires that baselines begin at the same point in time and that three consecutive baseline sessions be collected prior to the introduction of the intervention for each participant. Following a stable baseline, intervention data are collected continuously for the duration of treatment sessions. Coaching sessions were used to deliver the intervention, whereas homework sessions and observation sessions provided two contexts for weekly data collection. Visual analysis of the graphed data was used to examine changes in the dependent variables. For the first research question, the frequency of parent and child story-related utterances were the dependent variables. For the second research question, the frequency with which mothers used the three targeted intervention strategies (recasting, wh-questions, Intonation Prompts) were the dependent variables. For the third research question, child lexical diversity (i.e., number of

different vocabulary words) and mean length of utterance in morphemes (MLU) were the dependent variables. Generalized effects of the intervention were evaluated by examining lexical diversity and mean length of utterance produced during narrative language sampling at the post-treatment visit. Coaching sessions were coded for fidelity of intervention delivery.

### Structure of the Intervention Program

The intervention program for each dyad consisted of six types of activities: (a) baseline sessions, (b) parent education sessions, (c) weekly coaching sessions, (d) weekly homework/data collection sessions, (e) weekly clinician feedback sessions, and (f) weekly observation/data collection sessions. The sequence of sessions (c) through (f) (i.e., coaching, homework/data collection, clinician feedback, and observation/data collection) was maintained during each week of the intervention. Two individuals with training in speech/language pathology delivered the intervention. The primary interventionist was a licensed speech/language pathologist (i.e., SLP). The SLP was assisted by a speech/language clinician (i.e., Clinician) who had a masters degree in communication disorders and was completing a clinical fellowship year under the supervision of the SLP, according to the guidelines specified by the American Speech-Language Hearing Association ([www.asha.org](http://www.asha.org)). The SLP delivered the parent education and weekly coaching sessions. The Clinician reviewed the video-recordings of the homework sessions and provided the weekly feedback sessions to the mother. The clinician also emailed a written summary of the feedback session to the mother. The SLP and Clinician also watched the observation/data collection session. Each of the six intervention session types is explained in more detail below.

**Book Selection**—Following the pre-intervention visit, which was conducted onsite at the research clinic, each mother was invited to look through a collection of approximately 30 wordless picture books and select 9 books for use during the intervention. Digitized versions of the selected books were uploaded to the iPad. Digitized books were arbitrarily divided into 3 sets, with each set used for 4 consecutive weeks during the intervention. Thus, a new set of books was introduced during Intervention Weeks 1 (A<sub>1</sub>, B<sub>1</sub>, C<sub>1</sub>), 5 (A<sub>2</sub>, B<sub>2</sub>, C<sub>2</sub>) and 9 (A<sub>3</sub>, B<sub>3</sub>, C<sub>3</sub>). Immediately prior to each week's coaching session, the child selected a book from the designated set. Each book was placed back into its set as a potential choice after it was used for a complete week of intervention activities. Given that the child selected the book each week, a given book might not be selected at all or might be chosen up to three times over the course of the four-week book rotation. The books that were used by each dyad are listed in Table 2.

**Antecedent Behavior Supports**—Antecedent behavior supports were put in place to decrease the likelihood that problem behaviors would occur during the book-sharing activities (Sagai, Horner, & Sprague, 1999). To aid in the selection of appropriate antecedent behavior supports, a Board Certified Behavior Analyst (BCBA) evaluated each boy using: (a) Functional Behavioral Assessment Questionnaire (FBAQ; O'Neil, Horner, Albin, Storey, & Sprague, 1990); (b) Questions About Behavioral Function (QABF; Paclawskyj, Matsen, Rush, Smalls, & Vllmer, 2000); and (c) Functional Analysis Screening Tool (FAST; Florida Center on Self Injury, 2005). As all participants were reported to

struggle with task demands and actively engaging in non-preferred tasks, the primary support consisted of identifying a highly preferred reinforcer that could be made available contingent on completing the book sharing activity. Additionally, a token economy was implemented to assist participants in working for the delayed reinforcer. During clinician coaching sessions, tokens paired with verbal praise were delivered intermittently following a positive spoken or behavioral response during the session. When needed during coaching sessions, the clinician prompted the mother to deliver a token/praise. Additionally, clinician coaching and observation/data collection sessions were implemented at roughly the same day and time each week, thus helping to establish the predictability of the book-sharing activity for each dyad. Mothers were provided with a laminated icon representing each book, a choice board, a visual schedule depicting the format of the observation/data collection session, and a token tower with tokens. In addition to prompting and reinforcing the use of antecedent behavior support strategies during clinician coaching sessions, mothers were encouraged to use these strategies during homework/data collection and clinician observation/data collection sessions.

**Baseline sessions**—Baseline sessions were conducted via Skype™ and digitally captured for later transcription and coding. For each baseline session, the child selected one of the 9 books that made up the book set for that dyad. The mother was instructed to talk about the book in its entirety during each session. None of the books was viewed more than once during baseline and no instructions were provided to the mother except to look at the book with her child as she usually would. Dyads participated in 5, 7, and 8 baseline sessions, respectively, according to the logic of the multiple probe design. For each dyad, a stable baseline was achieved for each dependent variable prior to delivery of parent education sessions.

**Parent education sessions**—Following completion of baseline sessions for a given dyad, the SLP delivered an individualized powerpoint presentation containing information about the format and content of the intervention to each mother using the shared desktop application of Skype™. As a precaution against fatigue, the presentation was divided into two 45-min sessions that were delivered, at the mother's convenience, during the same week. During presentation of the parent education sessions, the SLP engaged each mother in discussion of the material and answered questions as they arose. Mothers also had access to an electronic copy of the powerpoint during the course of the intervention. Following the completion of the parent education sessions, each mother received an electronic copy of a written script for each of the 9 books selected for use during intervention activities. The script was intended as a guide to the sequence of story events and to highlight the vocabulary and grammatical constructions that could be used to tell each story. The mothers did depend on the use of the script during when the books were first introduced. Although this was not systematically studied, our impression is that mothers did not use the script once they were familiar with the story.

**Coaching sessions**—A coaching session, delivered by the SLP, was the first step in the weekly sequence of intervention-related activities. Each coaching session was conducted via Skype™, digitally captured, and coded to measure fidelity of intervention delivery. During

the coaching session, the mother and child looked at and talked together about the book selected for that week of the intervention. The SLP guided the mother's responses by modeling story-related vocabulary and sentence structure, by suggesting or modeling the use of the three targeted intervention strategies, and by reinforcing the mother's independent use of the intervention strategies. The SLP also provided cues for transitioning to the next page of the book and prompted or reinforced the mother as she shaped her child's on-task behavior using the token economy. During coaching, the mother wore a Bluetooth earpiece so that the SLP could interact with her without distracting the child. She was instructed to wear the earpiece on the side of her head that was facing away from the child. Because the text had been removed from the digitized books, the mother was encouraged to use the written script as needed as a guide for what she might say. Coaching sessions were not transcribed or coded to derive the variables representing parent and child story-related talking or child lexical diversity or MLU.

**Fidelity of clinician coaching**—Coaching sessions were coded from videotape to evaluate fidelity of intervention delivery. Two trained observers, who were not involved in delivering the intervention, independently coded each coaching session for the frequency of occurrence of coaching behaviors used by the SLP. Disagreements were resolved using consensus discussions. Agreement was calculated by using the formula: number of agreements/(number of agreements + disagreements) multiplied by 100. For each dyad, agreement was averaged within the following 5 categories of coaching behavior: (1) General coaching behaviors (e.g., providing a verbal model of a story-related utterances, providing the mother with a verbal model reinforcing a child utterance; providing reinforcement for a parent utterance, providing the mother with a prompt to transition to the next page of the book); (2) Recasts (verbal prompt for mother to use a recast; verbal model of a recast; verbal reinforcement of mother for using a recast); (3) Wh-questions (verbal prompt for mother to use a wh-question; verbal model of a wh-question; verbal reinforcement of mother for using a wh-question); (4) Intonation prompt (verbal prompt for mother to use an intonation prompt; verbal model of an intonation prompt; verbal reinforcement for using an intonation prompt); (5) Behavior management (prompt for mother to provide child with verbal praise; prompt for mother to provide token; verbal praise for mother's use of token). Means and ranges for each category of coaching behaviors, mean length of coaching sessions, and interobserver agreement for each dyad are summarized in Table 3.

**Independent homework/data collection sessions**—The homework session was the second step in the sequence of weekly intervention activities and provided a context in which to measure the mother's independent use of the strategies targeted during coaching sessions as well as the variables representing the child's spoken language (i.e., lexical diversity and MLU). Within four days of each coaching session, the mother was expected to share and discuss that week's story with her child, record the interaction using the Photobooth application on the iPad, and subsequently, upload that recording to the clinical team using Dropbox™. The mother was asked to read through the entire book with her child, just as the dyad had practiced during the coaching session, and she was allowed to use the written script if desired. The independent homework session could be completed at the mother's discretion as long as the Clinician would have sufficient time to watch the video

and provide feedback to the mother prior to the observation/data collection session for that week. Homework/data collection sessions were transcribed and coded to derive the frequency of parent and child story-related utterances and the frequency of maternal use of the targeted intervention strategies. Child lexical diversity and MLU were generated for by the transcription program.

**Clinician feedback sessions**—The Clinician reviewed the homework submitted by the mother and used this digitized video-recording to create clips illustrating parent use of the targeted intervention strategies. The clinician then participated in a SKYPE™ call with the mother and, using the video-clips or a brief transcription of various segments of the homework session, provided the mother with feedback about her performance and the child's responses, answered any questions posed by the mother, and engaged in joint problem solving relative to the child's behavior and spoken language. Clinician comments were also summarized in writing and sent to the mother electronically following each feedback session.

**Observation/data collection sessions**—The final step in the sequence of weekly intervention activities consisted of the SLP and Clinician observing, via SKYPE™, as the mother-child dyad engaged in the shared book reading activity. This session was digitally captured and provided a context, in addition to the homework/data collection session, from which to measure the mother's use of targeted intervention strategies as well as the variables representing the child's spoken language (i.e., lexical diversity and MLU). The difference between the independent homework/data collection session and the observation/data collection session was that, in the latter, the SLP and Clinician were online and actively watching the session as it occurred in real time. Although the SLP and the Clinician observed and digitally recorded this session, they did not provide any feedback, coaching, or guidance to the mother.

To limit the number of times during the week that the dyad was required to be involved in a SKYPE™ call, data for observation sessions was collected immediately prior to, and during the same SKYPE™ call as, the coaching session for the subsequent week/book. For example, observation/data collection for week 1 of the intervention was collected immediately prior to the coaching session for week 2 of the intervention, etc. In other words, the observation/data collection session represented the final intervention activity for a given week while the immediately-following coaching session represented the initial intervention activity for the subsequent week.

During the observation/data collection session, the mother was asked to share that week's selected book with her child and was allowed to use the written script if desired. The only substantive difference between the homework/data collection sessions and the observation/data collection sessions was that the mother was free to initiate the homework session with her child at her own convenience, whereas the time selected for the observation sessions was prearranged and generally did not vary from week to week. Observation/data collection sessions were transcribed and coded to derive the frequency of parent and child story-related utterances and the frequency of maternal use of the targeted intervention strategies. Child lexical diversity and MLU were generated for these sessions by the transcription program.

## Transcription, Coding, and Interobserver Agreement

Digitized video-recordings of baseline, homework/data collection and observation/data collection sessions were (a) transcribed using SALT (Systematic Analysis of Language Transcripts; Miller & Iglesias, 2008), a software program that enables the systematic transcription of samples of spoken language. Transcription was completed by highly trained research assistants following the procedures described by Abbeduto, Benson, Short, & Dolish (1995). Transcription involves a first draft by a primary transcriber, feedback from a second transcriber, and final editing by the primary transcriber. Use of this process averages >90% inter-observer agreement (Kover & Abbeduto, 2012). Finalized SALT transcripts were used to generate a set of standard measures for both speakers, including: total number of utterances (TNU), total number of words (TNW), number of different words (NDW), mean length of utterance in morphemes (MLU), number of filled pauses (mazes), and total number of unintelligible utterances. Finalized SALT transcripts also were coded and analyzed to generate the following variables:

**Maternal story-related talking**—Maternal utterances that directly related to the semantic or conceptual content of the story included: (a) models of story-related vocabulary and grammar (e.g., “The duck is thinking about getting a flashlight.”), (b) general story prompts (e.g., “How does the story start?”), (c) utterances that signaled transitions (e.g., “Let’s turn the page and see where the penguin will go next.”), and d) use of a targeted intervention strategy (i.e., WH-questions, Recasts, Intonation Prompts). The following types of utterances were excluded from the count of maternal story-related utterances: (a) utterances judged to be unrelated to the story; (b) requests for labels and sound effects; (c) yes/no questions; (d) questions without interrogative reversals (e.g., “The duck did what?”); (e) choice questions (e.g., “Who was driving -- the farmer or the goat?”); (f) utterances related to behavior management (e.g., “Put your hands down”); and (g) maternal responses that consisted entirely of filled pauses, false starts, repetitions, reformulations, or interjections.

**Child story-related talking**—Child utterances which directly related to the semantic or conceptual content of the study were coded from the finalized SALT transcripts. This variable utilized the SALT variable generated for total child utterances and excluded child utterances that: (a) were repetitions of a previous utterance referring to the same page of the book; (b) were completely unintelligible; or (c) that consisted entirely of a filled pause.

**Maternal use of WH-questions**—Maternal story-related utterances that were in the form of a WH-question were coded to yield the frequency of WH-questions. WH-questions that were simple requests for labels were not included in the count of WH-questions.

**Maternal use of Recasts**—Maternal story-related utterances that were contingent upon and related to the meaning of the immediately preceding child utterance and which provided additional or corrective semantic or grammatical content were coded as Recasts.

**Maternal use of Intonation Prompts**—Maternal story-related utterances that provided a partial utterance ending with an expectant pause were coded as Intonation Prompts.

**Interobserver agreement**—Using the completed SALT transcription, the Clinician completed all primary coding and one of three additional trained observers independently coded each homework and observation session for interobserver agreement. Transcripts used for interobserver agreement were randomly selected and assigned. For each transcript used for interobserver agreement, the records for the 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. Discrepancies were resolved by consensus discussion. Interobserver agreement for individually coded variables is presented in Table 4.

## Results

### Intervention Effects on Story-Related Talking

Intervention effects on parent and child story-related talking are presented in Figure 1 for Allan, Sam, Jay, and their mothers. Parent and child use of story-related talking increased for each of the three dyads. For Allan, low and decreasing levels of story-related talking occurred during baseline ( $M=48$  story-related utterances). His mother engaged in stable and very low levels of story-related talking during baseline ( $M=7$  story-related utterances). Upon introduction of the intervention, maternal use of story-related talking immediately increased in level ( $M=146$  utterances for the first 3 intervention sessions) and remained higher than baseline levels with no overlap throughout intervention ( $M=147$  story-related utterances). Upon introduction of the intervention, Allan's use of story-related talking immediately increased in level ( $M=213$  utterances) and exhibited a slight upward trend. Allan produced more story-related utterances than his mother during all sessions. Both maternal and child use of story-related talking was observed to occur with moderate variability during intervention. Prior to Session 8, both Allan and his mother produced more story-related utterances during homework sessions than during data collection sessions. Throughout the intervention, the duration of the parent-child interaction was longer during homework and observation sessions ( $M_s=17:20$  and  $16:24$  mins:secs, respectively) relative to baseline sessions ( $M=3:56$  min:secs).

For Sam, low levels of story-related talking occurred during baseline ( $M=42$  story-related utterances). His mother engaged in stable, low levels of story-related talking during baseline ( $M=59$  story-related utterances). Upon introduction of the intervention, maternal use of story-related talking immediately increased in level ( $M=145$  story-related utterances across the first 3 intervention sessions), exhibited an upward trend following intervention session 8, and showed minimal overlap (i.e., during one homework session) with the baseline phase. Upon introduction of the intervention, Sam's use of story-related talking immediately increased in level ( $M=147$  story-related utterances across the first 3 sessions;  $M=132$  story-related utterances across all intervention sessions), exhibited an upward trend following treatment session 8, and showed a minimal amount of overlap (i.e., during one observation session) with the baseline phase. Sam produced fewer story-related utterances than his mother during the majority of sessions. Both maternal and child use of story-related talking was observed to occur with some variability during intervention. For Sam and his mother, the duration of the parent-child interaction was longer during both homework and

observation sessions ( $M_s = 12:17$  and  $14:37$  mins:secs, respectively) relative to baseline sessions ( $M = 5:06$  min:secs).

Finally, Jay and his mother had a relatively flat, low baseline ( $M_s = 25$  and  $36$  story-related utterances, respectively), and both showed an immediate increase in level of talking ( $M_s = 103$  and  $136$  story-related utterances, respectively) when the intervention was introduced. During intervention, frequency of story-related talking by Jay and his mother did not overlap with the baseline phase. However, Jay's mother engaged in a higher frequency of story-related talking than did Jay. For this dyad, the duration of the parent-child interaction was longer during both homework and observation sessions ( $M_s = 12:09$  and  $14:26$  mins:secs, respectively) relative to baseline sessions ( $M = 3:42$  min:secs).

### Maternal Use of Targeted Intervention Strategies

Results for maternal use of WH-questions are presented in Figure 2 for all mothers. Allan's mother engaged in consistently low to zero frequency of WH-questions during baseline sessions and demonstrated an immediate increase in the level of this targeted strategy when the intervention was introduced. Overall, her use of WH-questions increased relative to baseline ( $M = 31$  WH-questions), but was highly variable (Range =  $10 - 66$  WH-questions). Specifically, during weeks 4 and 5 of the intervention, her use of WH-questions decreased in frequency. Two factors may account for this pattern. First, a substitute clinician conducted the coaching session during week 4 and her levels of coaching behavior were lower than in previous or subsequent sessions. Secondly, this clinician advised Allan's mother to focus on the use of Recasting during week 4, which may have influenced her use of WH-questions and Intonation Prompts. Subsequent to week 5, use of WH-questions by Allan's mother steeply increased to a high of 34 and 30 WH-questions, respectively, during weeks 9 and 10 of the intervention. The frequency of WH-questions then decreased to just over 24 WH-questions per session and remained relatively stable with a slight decreasing trend for the remainder of the intervention. Maternal use of WH-questions remained above baseline levels throughout the intervention phase.

During baseline, Sam's mother use WH-questions at a low level ( $M = 10$  WH-questions). Upon introduction of the intervention, she showed an immediate increase in the level of WH-questions ( $M = 63$  WH-questions during weeks 1 and 2 of the intervention) and, although her use of WH-questions was moderately variable (Range =  $27$  to  $73$  WH-questions), only the frequency of WH-questions during Homework Session 8 overlapped with baseline levels.

Jay's mother consistently displayed a very low to zero level use of WH-questions during baseline sessions, followed by a small but immediate increase in the level of use of this strategy following the introduction of the intervention. Although the level of WH-question use was moderately variable (Range =  $10$  to  $69$  WH-questions), the level of the data maintained a generally upward trend for the remainder of the intervention ( $M = 36$  WH-questions) and showed no overlap with baseline levels.

Results for maternal use of Recasts are presented in Figure 3. All of the mothers showed very low to zero level frequency of Recasts during baseline ( $M = 1, 8,$  and  $7$  Recasts,



respectively). Upon the introduction of the intervention, Allan's mother showed an immediate, moderate increase in use of Recasts, which she maintained above baseline levels throughout the remainder of the intervention ( $M = 37$  Recasts). Her use of Recasts across sessions was highly variable (Range = 17 – 74 Recasts).

Upon the introduction of intervention, Sam's mother showed an immediate, large increase in use of Recasts, which she maintained above baseline levels throughout the remainder of the intervention ( $M = 48$  Recasts), with little variability across sessions and an overall slight increasing trend.

Upon the introduction of intervention, Jay's mother showed an immediate, moderate increase in use of Recasts ( $M = 31$  Recasts) across the first 2 weeks of the intervention), which she maintained above baseline levels throughout the remainder of the intervention. Her use of Recasts was highly variable (Range = 19 – 91 Recasts), with an increasing trend followed by a slight decreasing trend. Use of Recasts remained above baseline levels throughout the intervention ( $M = 51$  Recasts), with zero overlap between baseline and intervention phases.

Results for maternal use of Intonation Prompts are presented in Figure 4. During baseline, Allan's mother engaged in low to zero levels of Intonation Prompts ( $M = 1$  Prompt, Range = 0 – 5 prompts). Upon introduction of the intervention, her use of Intonation Prompts showed a small but immediate increase ( $M = 8$  Prompts) through week 4 of the intervention. A larger, delayed effect was observed beginning with week 7 of the intervention. Following week 9, use of Intonation Prompts continued at above baseline levels with moderate variability (Range = 12 – 31 Prompts). Data for two homework sessions and one observation session during the first four weeks of the intervention overlapped with the first baseline data point.

During baseline, Sam's mother engaged in low frequency use of Intonation Prompts ( $M = 6$  prompts). An immediacy effect was not observed for Intonation Prompts upon introduction of the intervention. A delayed effect was observed beginning with a change in level at week 12 of the intervention and continued with a slight increasing trend through the remainder of the intervention. Fifty percent of the intervention data overlapped with the baseline data indicating that Sam's mother did not reliably acquire the spontaneous use of Intonation Prompts.

During baseline, Jay's mother engaged in low frequency use of Intonation Prompts ( $M = 4$  prompts). A large immediacy effect was observed upon introduction of the intervention. However, her use of Intonation Prompts temporarily decreased through week 4 of the intervention. Beginning with week 5 of the intervention, her use of Intonation Prompts increased and remained above baseline levels through the remainder of the intervention ( $M = 24$  prompts). During weeks 2, 3, and 4 of the intervention, data points for two homework and two observation sessions overlapped with the baseline data.

## Intervention Effects on Lexical Diversity and Grammatical Complexity

Intervention effects on lexical diversity and grammatical complexity are presented in Figure 5. In order to control for (a) duplications in the vocabulary words produced within baseline, homework, and observation sessions and (b) the differing numbers of books used during baseline and intervention sessions, we tallied the number of unique vocabulary words that were used within each session type (i.e., each different exemplar of a word was counted only once during baseline, homework or observation sessions). This sum was divided by the number of different books used during that phase of the intervention to yield a metric of unique vocabulary use per phase. Relative to baseline, all three boys increased the overall number of unique different vocabulary words they used during homework and observation sessions. Grammatical complexity was indexed through the use of mean length of utterance in morphemes and was generated by the SALT program. Sam and Jay, but not Allan, also increased the mean length of the utterances they produced during intervention sessions

Generalized effects of the intervention on child lexical diversity and grammatical complexity were measured during the narrative language sampling procedures and are presented in Table 5. Relative to the pre-intervention assessment, Sam and Jay increased the number of total words, total utterances, number of different words, and mean length of utterance produced in the narrative language sample with their mothers. Allan had a large increase in total words and total number of utterances but only a slight increase in number of different words and a decrease utterance length. During the narrative language sample with an unfamiliar examiner, Allan and Sam increased the number of total words, total utterances, number of different words, and length of utterances they produced relative to baseline. Jay, however, had a post-intervention language sample with the unfamiliar examiner that was almost three minute shorter than the pre-intervention sample. As a result of this, the number of total utterances, total number of words, and number of different words that he produced were fewer than at the pre-intervention period. His MLU was approximately the across the two time points. It is important to note that the unfamiliar examiner did not use any of the targeted intervention strategies when implementing the narrative procedure and, in fact, used a style of interaction that was designed to minimize the examiner's contribution to the interaction.

## Parent Satisfaction

All three mothers participated in a debriefing with the SLP and Clinician at the conclusion of the post-intervention visit to elicit feedback that could be used to modify the intervention in subsequent studies. Among other things, mothers were asked to describe their overall opinion about participation in the intervention, how easy or difficult it had been for them to learn the targeted strategies, their degree of comfort with the coaching process and the technology that had been used to deliver the intervention, whether they used the strategies at times during the day other than shared book reading, and whether they had shared the strategies with other individuals who interact with their child (i.e., teachers and therapists at school). Mothers reported a high degree of satisfaction with the intervention content and each mother independently reported to us that school-based professionals had noticed an improvement in their child's spoken language during the intervention. Each of the mothers also indicated that they: (a) were surprised at how long the boys were able to remain

engaged in the book sharing activities; and (b) were using the targeted intervention strategies outside of the treatment sessions

## Discussion

The goal of the current study was to examine a language intervention that targeted lexical diversity and grammatical complexity while scaffolding conversational turn-taking and topic maintenance for adolescent boys with FXS. The intervention was situated in shared storytelling, a treatment context that has frequently been utilized for younger children with a range of impairments affecting spoken language. Indeed, shared book reading has been termed a prototypical and iconic context for providing children with developmentally sensitive opportunities to learn language (Whitehurst & Lonigan, 1998). Clinician coaching was used to teach three evidence-based language support strategies to participating mothers with the goal of increasing the quality of verbal language input provided to their children during intervention sessions. Mothers were also encouraged to use a set of antecedent behavior support strategies to decrease the likelihood that child challenging behaviors would interfere with the book-sharing activity. We reasoned that the combined use of a structured intervention context, targeted language support strategies, and antecedent behavior supports would increase the length of time children could remain actively engaged in a conversational interaction and, thus, continue their exposure to enriched verbal language input (i.e., diverse vocabulary and more advanced sentence structure). The intervention was delivered through the use of distance video-teleconferencing, which has the potential to reach families who might otherwise not have access to intervention services. As a result of the intervention, all three boys demonstrated increases in lexical diversity and two of the three boys demonstrated increases in grammatical complexity during shared story telling activities with their mothers. Two of the three boys demonstrated generalized improvements in both lexical diversity and grammatical complexity. To our knowledge, this is the first study to directly evaluate the results of a spoken language intervention for adolescent males with FXS.

During the intervention sessions, each mother was asked to maintain the conversational interaction with her child until they had jointly looked at all of the pages of a selected book. Thus, the length of the conversational interaction, during which data was collected, was determined by the time it took to share a book from beginning to end. Relative to baseline sessions, all three dyads increased the amount of time they spent jointly engaged and the amount of story-related talking that was produced during each shared book-reading interaction. Behavioral engagement, with numerous embedded opportunities for child responses (Sutherland & Wehby, 2001), can be considered an index of active participation in learning activities. Active engagement provides a platform for early literacy learning and is considered to be crucial for later academic achievement, self-regulation, and social-emotional development (Hughes & Kwok, 2006). While most research examining the longitudinal effects of children's active engagement has involved preschool-aged children enrolled in classrooms (c.f., McWilliam, Trivette, & Dunst, 1985), the same facilitative effects of active engagement should be observed as a result of the sustained interaction between a verbally responsive parent and her child with an intellectual disability; that is, verbally responsive parenting practices should moderate the effects of child engagement on positive child language outcomes (Liew, Chen, & Hughes, 2010; Williford, Maier, Downer,

Pianta, & Howes, 2013). Establishing a routine context within which a child can be actively engaged in the learning activity should be particularly valuable to children with FXS who are known to have attentional challenges.

In general, increases in story-related talking were observed for both members of each dyad during homework sessions, which were independently initiated and recorded by the mother, as well as during data collection sessions, which were initiated, observed, and recorded by the SLP and Clinician. On average, the duration of time spent engaged in during observation sessions was longer than the duration of time spent engaged during homework sessions for two of the three mothers. This finding suggests that the presence of the clinician, even when no active coaching was involved, may have encouraged the mother to spend more time scaffolding the interaction with her child.

It is important to note that, not only was the duration of engagement in the book sharing activity increased, but both members of each dyad also increased the absolute frequency of story-related talking that was produced. Thus, during the shared story-book interaction, mothers modeled more diverse vocabulary words and more advanced grammatical structures for their children, provided verbally responsive language input by using Recasts, and helped to maintain the shared topic by using WH-questions. Similarly, mothers encouraged child participation and the child's use of newly introduced vocabulary words by providing Intonation Prompts. Taken together, the data suggest that participation in the intervention increased both the quality and quantity of verbal language input the children received. Such findings support the importance of active child engagement in the book-sharing activity accompanied by increases in maternal responsiveness (Kaderavek, Pentimonti, & Justice, 2014). The behaviors of mothers during the shared book-reading sessions incorporated four key aspects of responsivity: (a) contingent responding; (b) affective support; (c) following into the child's attentional focus; and, (d) rich language input (Landry & Smith, 2008).

The three intervention strategies that were taught the mothers are similar to some of the prompts and responding techniques used in dialogic reading, a well-established method that uses shared story-book reading to support language growth in young children (Justice & Pullen, 2003). In general, strategies used in dialogic reading programs include: providing numerous opportunities for the child to respond; asking children open-ended questions about story characters, settings, and events; expanding on children's answers; correcting in accurate responses; providing praise and encouragement; and, building on children's interests (Morgan & Meier, 2008). In the current study, we also included the use of Intonation Prompts which provides a grammatical frame to elicit a known vocabulary word (Bradshaw et al., 1998). In terms of the mother's acquisition of the three intervention strategies, all three mothers increased their spontaneous use of the WH-questions, which functioned to elicit child utterances, and Recasts, which provided developmentally advanced language contingent on the child's previous utterances. Two of the three mothers also increased their spontaneous use of Intonation Prompts. It was our clinical impression that Intonation Prompts were the most difficult strategy to teach to the mothers. Additionally, this strategy had a much lower frequency of use than the other two strategies. Both WH-questions and Intonation Prompts functioned to elicit child utterances. WH-questions may have been easier for mothers to use given that asking questions was a strategy mothers were

already familiar with using during conversational interactions with their children. That is, mothers used many questions during baseline (although most of these were yes/no questions, choice questions, questions that did not include story content, e.g. “What is that?”, and poorly phrased questions, e.g. “The duck went where?”) and perhaps more easily shifted to the use of WH-questions. Intonation Prompts, on the other hand, are not typically used in every day conversation and needed to be acquired and practiced by the mothers. In examining the data, it appears that Sam’s mother was using WH-questions rather than Intonation Prompts to elicit responses from Sam. In fact, she often used a question word when attempting to produce an Intonation Prompt (e.g., “The turtle went *where?*”). After week 7 of the intervention, her use of Intonation Prompts began to increase and no longer overlapped with baseline levels. In future iterations of this intervention, additional exemplars of this strategy as well as increased opportunities to practice Intonation Prompts during coaching and feedback sessions may need to be provided.

Intervention effects on structural aspects of child spoken language were examined by measuring the number of different unique words children used as well as the complexity of their spoken utterances during baseline, independent homework and observation sessions. With regard to vocabulary, each of the three boys increased the number of unique vocabulary words produced when sharing each book during homework and observation sessions relative to baseline sessions. Thus, participation in the intervention provided each boy with regular opportunities to practice using a broader variety of spoken vocabulary words than they likely would have used without the intervention.

With regard to the complexity of the grammar produced during intervention sessions, two of the boys (Sam and Jay) showed an increase in mean length of utterance relative to baseline whereas one participant (Allan) showed a decrease. One potential explanation for the reduction in MLU that was noted for Allan is that his utterances during baseline often were grammatically correct but not semantically or conceptually meaningful. For example, when narrating the book *Cow Cant Sleep* during baseline, Allan said, “The cow liked to boom at an entire hole”. This utterance contains 10 morphemes but does not make conceptual sense. Similarly, Allan produced the utterance, “And the horse friend helps the pig get out his old coconuts”. Sentences such as these might have inflated Allan’s baseline MLU. Additionally, Allan frequently repeated the last several words of his mother’s utterances during treatment sessions. For example, when Allan’s mother said, “Harry felt sad because the family did not recognize him,” Allan repeated, “recognize him”. When Allan’s mother said, “They didn’t recognize Harry because he was so dirty”, Allan repeated “So dirty”. Brief repetitions such as these may have served to lower Allan’s treatment MLU. It should be noted that, in some of these overlapping utterances, Allan’s mother could have used an Intonation Prompt as Allan could potentially have completed the utterance independently.

Thus, lengthy but conceptually invalid utterances may have served to increase Allan’s MLU during baseline. Conversely, immediate partial imitations of his mother’s preceding utterances may have served to decrease Allan’s MLU during treatment sessions. To examine the former explanation, we coded Allan’s baseline, homework, and observation/data collection sessions to identify the proportion of child utterances which were conceptually inadequate. In support of our working hypothesis, coding revealed that, on average, over

21% of Allan's baseline utterances were semantically incongruous while less than 2% of utterances were semantically incongruous during treatment sessions. We then recalculated Allan's MLU using only semantically correct utterances and after removing utterances that were in direct or partial imitation of his mother. We also excluded Allan's responses to intonation prompts (which were likely to be a single word). There was still a difference, however, favoring baseline MLU (4.30 words during baseline vs 3.70 during treatment). It might be possible that the increased amount of talking by Allan's mother during treatment sessions decreased Allan's opportunities to produce longer utterances and this is something that should be carefully monitored in future studies. Additionally, it should be noted that Allan had fewer than 50 semantically correct utterances in four of the five baseline sessions resulting in a very limited sample of utterances from which to compute MLU. Thus, Allan's baseline MLU may not have been representative of his actual language ability. Some researchers do, in fact, suggest that MLU levels exceeding 3.0 are less closely associated with grammatical development relative to lower levels of MLU (Scarborough, Rescorla, Tager-Flusberg, Fowler, & Sudhalter, 1991); that is, utterances can include content words which inflate MLU without necessarily being grammatical (Eisenberg, Fersko, Lundgren, 2001). Regardless of the explanation, however, it can be concluded that the intervention had positive effects in the domain of expressive vocabulary and that Allan used more utterances that were semantically meaningful and related to the events in the story during treatment sessions.

During the intervention, mothers were taught to respond to semantically incongruous utterances by providing a corrective model, by using a wh-question to clarify Allan's meaning, or by using an intonation prompt to elicit an appropriate vocabulary word. The use of corrective feedback (which is also a part of dialogic reading interventions; Whitehurst et al., 1994) serves to highlight one of the advantages of using shared story-telling to structure a language intervention session for boys with FXS; that is, boys with FXS frequently use tangential and repetitive utterances during conversational interactions. Maintenance of a shared topic, scaffolded by an illustrated book, not only helped to constrain child responses and keep the conversation on track, but also provided an indication as to what semantic content the child might be trying to express. In this way, mothers were able to provide feedback to help their child more accurately express their intended meanings.

During the narrative language sample with their mothers at the post-intervention, all of the boys remained engaged in the task for a longer duration of time relative to the pre-intervention narrative and produced more total words and utterances. All three boys also produced a more diverse set of vocabulary words during the post-intervention sample, although improvement in the number of different words was not as striking as for Allan as for Sam and Jay. As was the case during intervention sessions, Allan's MLU was shorter in the post-intervention sample. In addition to examining the proximal effects of the intervention, narrative language sampling with an unfamiliar examiner was used at the pre- and post-intervention time points to provide a context for evaluating the generalized effects of the intervention on the boys' spoken language use. In the generalization context at the post-intervention assessment, two of the three boys (Allan and Sam) stayed engaged in the narrative task for almost two minutes longer than they had prior to the intervention. These boys produced more total words and utterances than they had prior to the intervention.

Additionally, they demonstrated a more diverse vocabulary and longer MLU relative to the sample with the unfamiliar examiner that was collected prior to the intervention. One interpretation of Jay's lack of generalized gains in vocabulary and mean length of utterance at the post-test may be that he was just in a hurry to complete the session and, therefore, did not stay engaged and perform at an acceptable level.

It is important to note the increased duration of time the boys were able to remain engaged in the narrative language sampling procedure at the post-intervention assessment, an increase that is particularly noteworthy when interacting with their mothers. As previously mentioned, boys with FXS have difficulty sustaining their attentional focus; a behavioral characteristic that almost certainly has a negative impact on the ability to stay on topic and engage in sustained conversational interactions. By increasing the amount of time that a child remains engaged in a conversation, parents can increase the amount of responsive verbal language the child receives. Over time, increasing child exposure to developmentally advanced models of vocabulary, grammar, and syntax, should have a positive impact on spoken language growth. Although it is clear that the mothers had learned to increase the duration of interaction with their children by using the three targeted intervention strategies as well as the antecedent behavior supports, it is also important to note that both Allan and Sam increased the duration of their narratives with an unfamiliar examiner who used the same interaction style as had been used during the pre-intervention narrative language sample.

### **Limitations and future directions**

Although these preliminary results are promising, there are several limitations that should be mentioned and many future directions to explore. One limitation involves the digitized books that were used during the intervention. These books varied in length from 14 to 30 pages; a discrepancy which almost certainly affected the duration of the book sharing interaction as well as the diversity of vocabulary words which were used. Future replications and extensions of this intervention approach should standardize the length of the books used.

The goal in designing the weekly structure of intervention activities was threefold. First, that the boys to have sufficient familiarity with each book used during the intervention to understand the gist of the story, to be able to predict the events in the story, and to be exposed to new vocabulary words. Second, to provide each mother with ample opportunities to practice the use of the three targeted intervention strategies and to incorporate the feedback received in real-time during clinician coaching sessions. Finally, that the clinicians to have the chance to provide the mother with additional feedback after practicing the book sharing activity independently (i.e., during homework). Thus, four intervention-related activities were delivered each week: clinician coaching, independent homework/data collection, clinician feedback, clinician observation/data collection. The clinician feedback session that followed the independent homework session only involved the mother and the clinician; the child did not participate in this session. These sessions tended to vary in length and their format changed over the course of the intervention. The clinician and the mother would typically watch and discuss video clips from the independent homework session, and the clinician would sometimes provide written feedback. It is possible that a more structured

session that incorporated written feedback, a review of the homework video, as well as periodic revisiting of the strategies and the rationale for how the strategies could enhance language input for the child would have provided the mothers with more confidence in their abilities and a greater understanding of their critical role in the intervention. Observation data collection for a given book was conducted immediately prior to, but during the same Skype session as, the clinician coaching session for the next book. This resulted in a session that lasted approximately 45 minutes which may have been too lengthy. Maternal uptake of the intervention and child compliance might have been improved if the clinician observation session and clinician coaching sessions were conducted on different days even though this would have meant that there was an intervention activity on an additional day each week.

Another limitation involves the number of times each dyad used a book. Mothers selected three sets of books consisting of three books each. A different book was used for each baseline session. However, each book set was used for 4 consecutive weeks during the intervention phase of the study, resulting in all of the dyads using one of the books in each of the book sets twice. This involved re-using a given book during coaching, independent homework, and clinician observation sessions such that, overall, these particular 3 books were used a total of 6 times during the intervention phase of the study. In planning the intervention, we had reasoned that some repetition of the books would be beneficial, in terms of repeated exposure to vocabulary and story structure. However, it was our clinical impression during the intervention that the boys were bored with this much repetition and that repeated exposure to the same book during sequential weeks of the intervention should be avoided. In upcoming iterations of the program, each set will be made up of 5 books and a new set of books will be used every four weeks. After a book is used, it will not be replaced in a set and the book that is unused after 4 weeks will rotate into the next book set. The use of 5 books will provide the child with a choice of two new books for each week of the intervention.

Despite having the content of the intervention available to them in powerpoint format on their borrowed laptop computer, none of the mothers reported that they had referred back to the powerpoint at any time during the intervention. Future iterations of this study should build in opportunities for the mothers to review the content of the parent education sessions at regular intervals, such as during the feedback sessions. A final limitation is that only mothers were enrolled in the current study. Because mothers of children with FXS are themselves carriers of the pre- or full mutation of the FMR1 gene, we sought to limit variability in our small sample by focusing this intervention on mothers. There would be no reason that fathers could not also be taught to use the targeted intervention strategies. In fact, the intervention effects might likely be stronger if both parents were involved in delivering the intervention as this would increase the likelihood that the child would be exposed to the intervention strategies throughout the time spent with their family.

This study provides preliminary evidence for the efficacy of an intervention aimed at improving the spoken language of adolescent boys with fragile X syndrome and delivered entirely through the use of distance teleconferencing technology. Because few language interventions have been evaluated for use with adolescents with intellectual and developmental disabilities, this study addresses an area of pressing need for teachers,



clinicians, and other professionals who work with individuals in this age range. By using a parent-mediated intervention model, the intervention was delivered to a family member who would be able to use the intervention regularly and who could generalize the use of intervention strategies to other daily interactions with the child. In addition, this intervention program could easily be adapted to be implemented in the classroom or resource room. Providing a narrative language intervention in a school-based setting could be useful given the strong relationship between narrative language and reading comprehension.

Although trajectories of change may be limited, previous research suggests that some aspects of language learning continue into adolescence for individuals with intellectual and developmental disabilities, including Down syndrome (e.g., Chapman, Hesketh, & Kistler, 2002) and FXS (Pierpont, Richmond, Abbeduto, Kover, & Brown, 2012). The current descriptive findings provide the impetus for examining behavioral interventions aimed at improving language growth in adolescents with FXS and other types of neurodevelopmental disorders. Overall, results of this pilot study suggest that a parent-implemented language intervention based on shared story telling can be an effective way to support spoken language for older school-aged boys with FXS. Mothers reported high levels of satisfaction with their participation in the intervention and with their children's progress. While our initial results are promising, a randomized group experiment will be needed to confirm the preliminary results reported here.

## Acknowledgements

We gratefully acknowledge the MIND Institute Intellectual and Developmental Disabilities Research Center funded by the Eunice Kennedy Shriver National Institute for Child Health and Human Development (U54 HD 079125, PI: Leonard Abbeduto) that enabled us to complete this research project. We extend our deepest appreciation to the families who participated in this research. Preliminary results from this project were presented in poster format at the 48th Annual Gatlinburg Conference on Research and Theory in Intellectual and Developmental Disabilities (April, 2015; New Orleans, LA).

## References

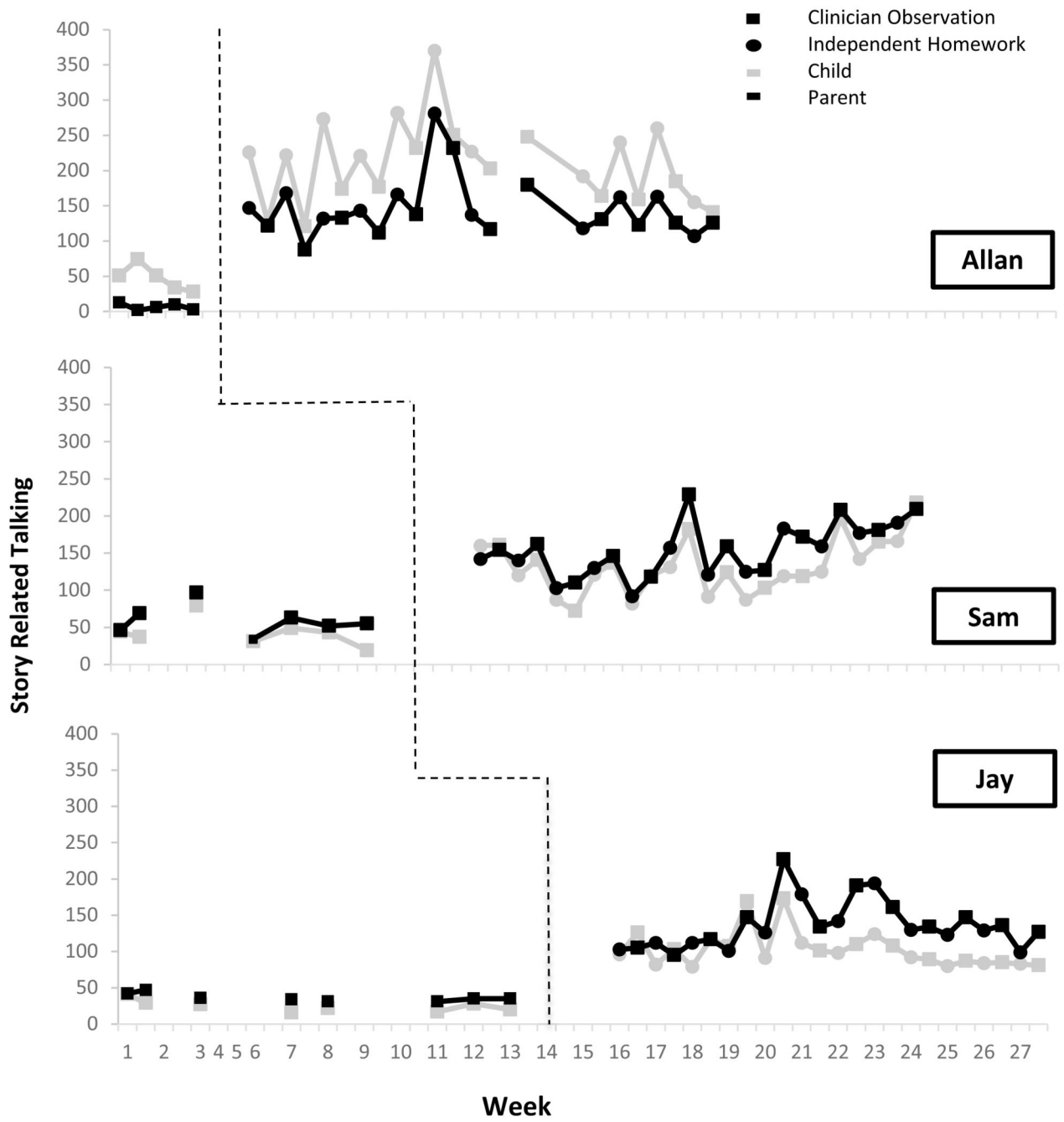
- Abbeduto L, Benson G, Short K, Dolish J. Effects of sampling context on the expressive language of children and adolescents with mental retardation. *Mental Retardation*. 1995; 33:279–288. [PubMed: 7476250]
- Abbeduto L, Brady N, Kover S. Language development and fragile X syndrome: Profiles, syndrome-specificity, and within-syndrome differences. *Mental Retardation and Developmental Disabilities Research Reviews*. 2007; 13:36–46. [PubMed: 17326110]
- Abbeduto L, Hagerman R. Language and communication in fragile X syndrome. *Mental Retardation and Developmental Disabilities Research Reviews*. 1997; 3:313–322.
- Albanese O, Antoniotti C. Teacher dialogue style and children's story comprehension. *European Journal of Psychology of Education*. 1997; 12:249–259.
- Bailey D, Mesibov G, Hatton D, Clark R, Roberts J, Mayhew L. Autistic behavior in young boys with fragile X syndrome. *Journal of Autism and Developmental Disorders*. 1998; 28:499–508. [PubMed: 9932236]
- Bellon-Harn M, Hoffman P, Harn W. Use of cloze and contrast word procedures in repeated storybook reading: Targeting multiple domains. *Journal of Communication Disorders*. 2004; 37:53–75. [PubMed: 15013379]
- Belser R, Sudhalter V. Conversational characteristics of children with fragile X syndrome: repetitive speech. *American Journal on Mental Retardation*. 2001; 106:28–38. [PubMed: 11246710]

- Bhakar A, Dölen G, Bear M. The pathophysiology of fragile X (and what it teaches us about synapses). *Annual Review of Neuroscience*. 2012; 35:417–443.
- Bradshaw M, Hoffman P, Norris J. Efficacy of expansions and cloze procedures in the development of interpretations by preschool children exhibiting delayed language development. *Language, Speech, and Hearing Services in Schools*. 1998; 29:85–95.
- Brady N, Warren S, Fleming K, Keller J, Sterling A. Effect of sustained maternal responsivity on later vocabulary development in children with fragile X syndrome. *Journal of Speech, Language, and Hearing Research*. 2014; 57:212–226.
- Bruner J. Vygotsky: A historical and conceptual perspective. *Culture, communication, and cognition: Vygotskian perspectives*. 1985:21–34.
- Brown, W. The molecular biology of the fragile X mutation. In: Hagerman, R.; Hagerman, P., editors. *Fragile X Syndrome: Diagnosis, Treatment and Research*. 3rd ed.. The Johns Hopkins University Press; Baltimore, MD: 2002. p. 110-135.
- Camarata, S. A rationale for naturalistic speech intelligibility intervention. In: Fey, M.; Windsor, J.; Warren, S., editors. *Language Intervention: Preschool through the Elementary Years*. Brookes Publishing Company; Baltimore, MD: 1995. p. 63-84.
- Chapman RS, Hesketh LJ, Kistler DJ. Predicting longitudinal change in language production and comprehension in individuals with down syndrome: Hierarchical linear modeling. *Journal of Speech, Language, and Hearing Research*. 2002; 45(5):902–915.
- Charlop M, Schreibman L, Thibodeau M. Increasing spontaneous verbal responding in autistic children using a time delay procedure. *Journal of Applied Behavior Analysis*. 1985; 18:155–166. [PubMed: 4019351]
- Cirrin F, Gillam R. Language intervention practices for school-age children with spoken language disorders: A systematic review. *Language, Speech, and Hearing Services in Schools*. 2008; 39:110–137.
- Cordeiro L, Ballinger E, Hagerman R, Hessl D. Clinical assessment of DSM-IV anxiety disorders in fragile X syndrome: Prevalence and characterization. *Journal of Neurodevelopmental Disorders*. 2011; 3:57–67. [PubMed: 21475730]
- Dale P, Crain-Thoreson C, Notari-Syverson A, Cole K. Parent-child storybook reading as an intervention technique for young children with language delays. *Topics in Early Childhood Special Education*. 1996; 16:213–235.
- Dickinson D, Hofer K, Barnes E, Grifenhagen J. Examining teachers' language in Head Start classrooms from a Systemic Linguistics Approach. *Early Childhood Research Quarterly*. 2014; 29:231–244.
- Dickinson D, Porche M. Relation between language experiences in preschool classrooms and children's kindergarten and fourth-grade language and reading abilities. *Child Development*. 2011; 82:870–886. [PubMed: 21413936]
- Dunst C, Williams A, Trivette C, Simkus A, Hamby D. Relationships between inferential book reading strategies and young children's language and literacy competence. *Center for Early Literacy Learning*. 2012; 5:1–10.
- Eisenberg SL, Fersko TM, Lundgren C. The use of MLU for identifying language impairment in preschool children: A review. *American Journal of Speech Language Pathology*. 2001; 10(4):323–342.
- Estigarribia B, Martin G, Roberts J. Cognitive, environmental, and linguistic predictors of syntax in fragile X syndrome and Down syndrome. *Journal of Speech, Language, and Hearing Research*. 2012; 55:1600–1612.
- Estigarribia B, Roberts J, Sideris J, Price J. Expressive morphosyntax in boys with fragile X syndrome with and without autism spectrum disorder. *International Journal of Language & Communication Disorders*. 2011; 46:216–230. [PubMed: 21401819]
- Fey, M. *Language intervention with young children*. College-Hill Press; Hillsborough, TX: 1986.
- Fey M, Cleave P, Ravida A, Long S, DeJmal A, Easton D. Effects of grammar facilitation on the phonological performance of children with speech and language impairments. *Journal of Speech, Language, and Hearing Research*. 1994; 37:594–607.

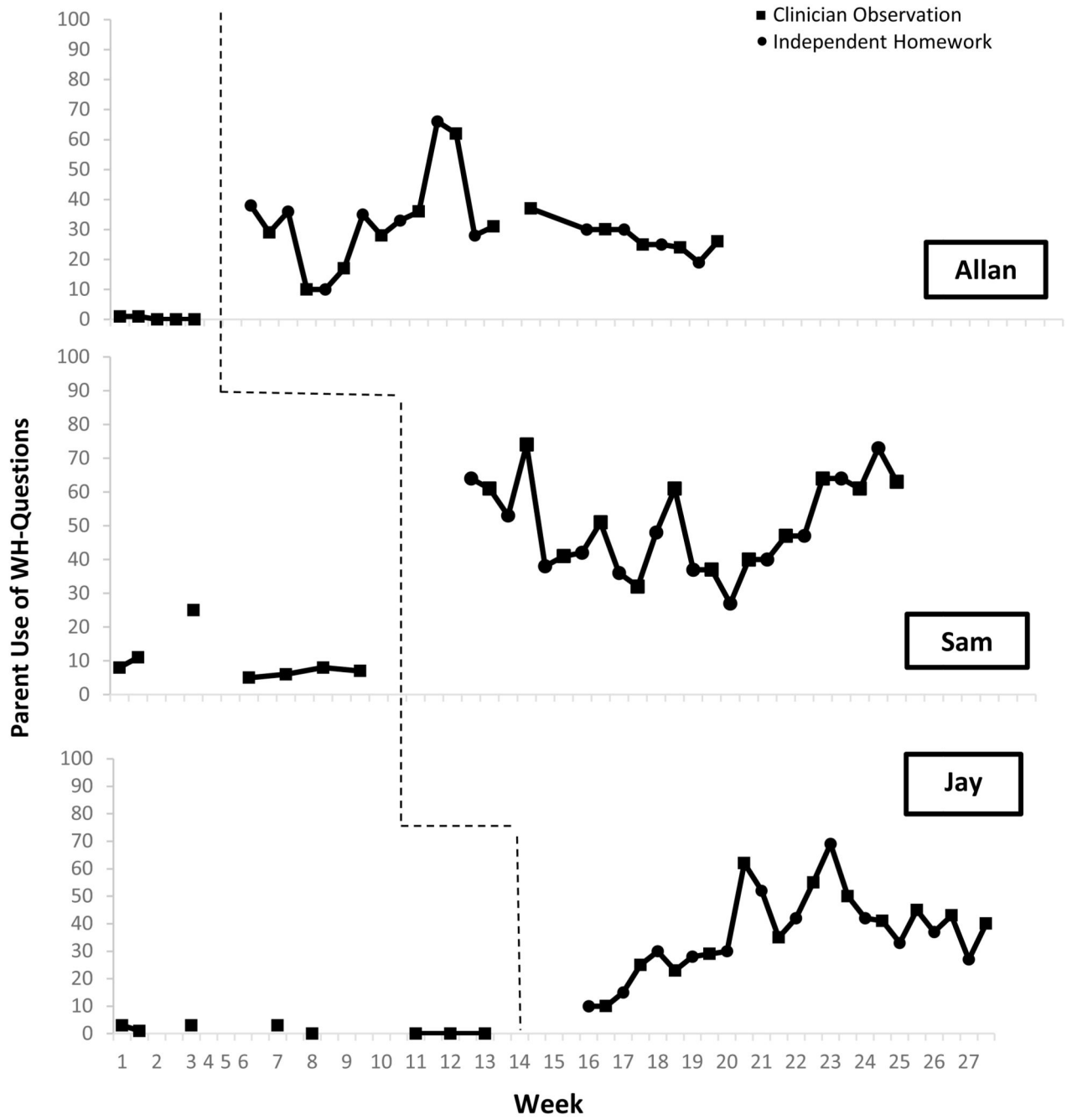
- Green L, Klecan-Aker J. Teaching story grammar components to increase oral narrative ability: A group intervention study. *Child Language Teaching and Therapy*. 2012; 28:263–276.
- Hall S, Lightbody A, Hirt M, Rezvani A, Reiss A. Autism in FXS: A category mistake? *Journal of the American Academy of Child & Adolescent Psychiatry*. 2010; 49:921–933. [PubMed: 20732628]
- Hartley S, Seltzer M, Raspa M, Olmstead M, Bishop E, Bailey D Jr. Exploring the adult life of men and women with FXS: Results from a national survey. *American Journal of Intellectual and Developmental Disabilities*. 2011; 116:16–35.
- Hauser-Cram P, Warfield M, Shonkoff J, Krauss M, Sayer A, Upshur C, Hodapp R. Children with disabilities: A longitudinal study of child development and parent well-being. *Monographs of the Society for Research in Child Development*. 2001:i–126. [PubMed: 11677873]
- Hessl D, Nguyen D, Green C, Chavez A, Tassone F, Hagerman R, Hall S. A solution to limitations of cognitive testing in children with intellectual disabilities: The case of FXS. *Journal of Neurodevelopmental Disorders*. 2009; 1:33–45. [PubMed: 19865612]
- Hogan T, Bridges M, Justice L, Cain K. Increasing higher level language skills to improve reading comprehension. *Focus on Exceptional Children*. 2011; 44:1–20.
- Iwata B, DeLeon I, Roscoe E. Reliability and validity of the functional analysis screening tool. *Journal of Applied Behavior Analysis*. 2013; 46:271–284. [PubMed: 24114099]
- Justice LM, Pullen PC. Promising interventions for promoting emergent literacy skills three evidence-based approaches. *Topics in Early Childhood Special Education*. 2003; 23(3):99–113.
- Kaderavek J, Justice L. Shared storybook reading as an intervention context: Practices and potential pitfalls. *American Journal of Speech-Language Pathology*. 2002; 11:395–406.
- Kaderavek JN, Pentimonti JM, Justice LM. Children with communication impairments: Caregivers' and teachers' shared book-reading quality and children's level of engagement. *Child Language Teaching and Therapy*. 2014; 30(3):289–302.
- Kaiser A, Roberts M. Parent-implemented enhanced milieu teaching with preschool children who have intellectual disabilities. *Journal of Speech, Language, and Hearing Research*. 2013; 56:295–309.
- Kover S, McDuffie A, Abbeduto L, Brown W. Effects of sampling context on spontaneous expressive language in males with fragile X syndrome or Down syndrome. *Journal of Speech, Language, and Hearing Research*. 2012; 55:1022–1038.
- Landry SH, Smith KE. Family processes that support school readiness: Specific behaviors and contextual conditions that set this process in motion. *Disparities in school readiness: How families contribute to transitions into school*. 2008:85–107.
- Leigh MJ, Nguyen D, Mu Y, Winarni TI, Schneider A, Chechi T, Hagerman R. A randomized double-blind, placebo-controlled trial of minocycline in children and adolescents with fragile X syndrome. *Journal of Developmental and Behavioral Pediatrics*. 2013; 34:147–155. [PubMed: 23572165]
- Levy Y, Gottesman R, Borochowitz Z, Frydman M, Sagi M. Language in boys with fragile X syndrome. *Journal of Child Language*. 2006; 33:125–144. [PubMed: 16566323]
- Liew J, Chen Q, Hughes JN. Child effortful control, teacher-student relationships, and achievement in academically at-risk children: Additive and interactive effects. *Early Childhood Research Quarterly*. 2010; 25(1):51–64. [PubMed: 20161421]
- Lord, C.; Rutter, M.; DiLavore, P.; Risi, S. *Autism diagnostic observation schedule: ADOS*. Western Psychological Services; Los Angeles, CA: 2002.
- Martin G, Losh M, Estigarribia B, Sideris J, Roberts J. Longitudinal profiles of expressive vocabulary, syntax and pragmatic language in boys with FXS or DS. *International Journal of Language & Communication Disorders*. 2013; 48:432–443. [PubMed: 23889838]
- McWilliam R, Trivette C, Dunst C. Behavior engagement as a measure of the efficacy of early intervention. *Analysis and Intervention in Developmental Disabilities*. 1985; 5(1):59–71.
- Merenstein S, Sobesky W, Taylor A, Riddle J, Tran H, Hagerman R. Molecular-clinical correlations in males with an expanded FMR1 mutation. *American Journal of Medical Genetics*. 1996; 64:388–394. [PubMed: 8844089]
- Miles S, Chapman R, Sindberg H. Sampling context affects MLU in the language of adolescents with Down syndrome. *Journal of Speech, Language, and Hearing Research*. 2006; 49:325–337.

- Miller J, Iglesias A. Systematic Analysis of Language Transcripts (SALT), English & Spanish (Version 9)[Computer software]. Madison: University of Wisconsin—Madison, Waisman Center. Language Analysis Laboratory. 2008
- Morgan PL, Meier CR. Dialogic reading's potential to improve children's emergent literacy skills and behavior. *Preventing School Failure: Alternative Education for Children and Youth*. 2008; 52(4): 11–16. [PubMed: 25960623]
- Moss J, Oliver C, Arron K, Burbidge C, Berg K. The prevalence and phenomenology of repetitive behavior in genetic syndromes. *Journal of Autism and Developmental Disorders*. 2009; 39:572–588. [PubMed: 19037716]
- O'Neill, RE.; Horner, RH.; Albin, RW.; Storey, K.; Sprague, JR. Functional analysis of problem behavior: A practical assessment guide. Sycamore Publishing Company; 1990.
- Oostra B, Willemsen R. FMR1: a gene with three faces. *Biochimica et Biophysica Acta (BBA)-General Subjects*. 2009; 1790(6):467–477. [PubMed: 19233246]
- Paclawskyj T, Matson J, Rush K, Smalls Y, Vollmer T. Questions about behavioral function (QABF): A behavioral checklist for functional assessment of aberrant behavior. *Research in Developmental Disabilities*. 2000; 21:223–229. [PubMed: 10939320]
- Pentimonti J, Zucker T, Justice L, Petscher Y, Piasta S, Kaderavek J. A standardized tool for assessing the quality of classroom-based shared reading: Systematic assessment of book reading (SABR). *Early Childhood Research Quarterly*. 2012; 27:512–528.
- Petersen D, Gillam S, Spencer T, Gillam R. The effects of literate narrative intervention on children with neurologically based language impairments: An early stage study. *Journal of Speech, Language, and Hearing Research*. 2010; 53:961–981.
- Petersen D. Systematic review of narrative-based language intervention with children who have language impairment. *Communication Disorders Quarterly*. 2011; 32:207–220.
- Pierpont EI, Richmond EK, Abbeduto L, Kover ST, Brown WT. Contributions of phonological and verbal working memory to language development in adolescents with fragile X syndrome. *Journal of neurodevelopmental disorders*. 2011; 3(4):335–347. [PubMed: 21993552]
- Pratt M, Kerig P, Cowan P, Cowan C. Mothers and fathers teaching 3-year-olds: Authoritative parenting and adult scaffolding of young children's learning. *Developmental Psychology*. 1988; 24:832.
- Price J, Roberts J, Hennon E, Berni M, Anderson K, Sideris J. Syntactic complexity during conversation of boys with fragile X syndrome and Down syndrome. *Journal of Speech, Language, and Hearing Research*. 2008; 51:3–15.
- Price J, Roberts J, Vandergrift N, Martin G. Language comprehension in boys with fragile X syndrome and boys with Down syndrome. *Journal of Intellectual Disability Research*. 2007; 51:318–326. [PubMed: 17326813]
- Roberts JE, Hennon EA, Price JR, Dear E, Anderson K, Vandergrift NA. Expressive language during conversational speech in boys with fragile X syndrome. *American Journal on Mental Retardation*. 2007; 112(1):1–17. [PubMed: 17181388]
- Roberts J, Mirrett P, Burchinal M. Receptive and expressive communication development of young males with fragile X syndrome. *American Journal on Mental Retardation*. 2001; 106:216–230. [PubMed: 11389664]
- Sugai G, Horner R, Sprague J. Functional-assessment-based behavior support planning: Research to practice to research. *Behavioral Disorders*. 1999:253–257.
- Sameroff A, Fiese B. Transactional regulation: The developmental ecology of early intervention. *Handbook of Early Childhood Intervention*. 2000; 2:135–159.
- Scarborough HS, Rescorla L, Tager-Flusberg H, Fowler AE, Sudhalter V. The relation of utterance length to grammatical complexity in normal and language-disordered groups. *Applied Psycholinguistics*. 1991; 12(01):23–46.
- Scerif G, Longhi E, Cole V, Karmiloff-Smith A, Cornish K. Attention across modalities as a longitudinal predictor of early outcomes: The case of fragile X syndrome. *Journal of Child Psychology and Psychiatry*. 2012; 53:641–650. [PubMed: 22211574]

- Schwartz R, Chapman K, Terrell B, Prelock P, Rowan L. Facilitating word combination in language-impaired children through discourse structure. *Journal of Speech and Hearing Disorders*. 1985; 50:31–39. [PubMed: 3974209]
- Snow, C.; Perlman, R.; Nathan, D. Why routines are different: Toward a multiple-factor model of the relationship between input and language acquisition. In: Nelson, K.; van Kleeck, A., editors. *Children's language*. Vol. 6. Lawrence Erlbaum Associates; Hillsdale, NJ: 1987. p. 65-97.
- Sutherland KS, Wehby JH. Exploring the relationship between increased opportunities to respond to academic requests and the academic and behavioral outcomes of students with EBD. *A review. Remedial and Special Education*. 2001; 22(2):113–121.
- Swanson L, Fey M, Mills C, Hood L. Use of narrative-based language intervention with children who have specific language impairment. *American Journal of Speech-Language Pathology*. 2005; 14:131–141. [PubMed: 15989388]
- Symons F, Clark R, Hatton D, Skinner M, Bailey D. Self-injurious behavior in young boys with Fragile X syndrome. *American Journal of Medical Genetics Part A*. 2003; 118:115–121. [PubMed: 12655491]
- Tyler A, Sandoval K. Preschoolers with phonological and language disorders Treating different linguistic domains. *Language, Speech, and Hearing Services in Schools*. 1994; 25(4):215–234.
- van Kleeck A, Vander Woude J, Hammett L. Fostering literal and inferential language skills in Head Start preschoolers with language impairment using scripted book-sharing discussions. *American Journal of Speech-Language Pathology*. 2006; 15:85–95. [PubMed: 16533095]
- Warren S, Brady N, Sterling A, Fleming K, Marquis J. Maternal responsivity predicts language development in young children with fragile X syndrome. *American Journal on Intellectual and Developmental Disabilities*. 2010; 115:54–75. [PubMed: 20025359]
- Warren S, Yoder P. Emerging model of communication and language intervention. *Mental Retardation and Developmental Disabilities Research Reviews*. 1997; 3:358–362.
- Whitehurst G, Arnold D, Epstein J, Angell A, Smith M, Fischel J. A picture book reading intervention in day care and home for children from low-income families. *Developmental Psychology*. 1994; 30:679.
- Whitehurst GJ, Lonigan CJ. Child development and emergent literacy. *Child Development*. 1998; 69(3):848–872. [PubMed: 9680688]
- Williford AP, Maier MF, Downer JT, Pianta RC, Howes C. Understanding how children's engagement and teachers' interactions combine to predict school readiness. *Journal of Applied Developmental Psychology*. 2013; 34:299–309. [PubMed: 26722137]
- Yoder P, Davies B, Bishop K, Munson L. Effect of adult continuing wh-questions on conversational participation in children with developmental disabilities. *Journal of Speech, Language, and Hearing Research*. 1994; 37:193–204.
- Yoder P, Spruytenburg H, Edwards A, Davies B. Effect of verbal routine contexts and expansions on gains in the mean length of utterance in children with developmental delays. *Language, Speech, Hearing Services in Schools*. 1995; 26:21–32.
- Zevenbergen A, Whitehurst G, Zevenbergen J. Effects of a shared-reading intervention on the inclusion of evaluative devices in narratives of children from low-income families. *Journal of Applied Developmental Psychology*. 2003; 24:1–15.
- Zucker T, Justice L, Piasta S, Kaderavek J. Preschool teachers' literal and inferential questions and children's responses during whole-class shared reading. *Early Childhood Research Quarterly*. 2010; 25:65–83.



**Figure 1.**  
Child and Parent Story Related Talking



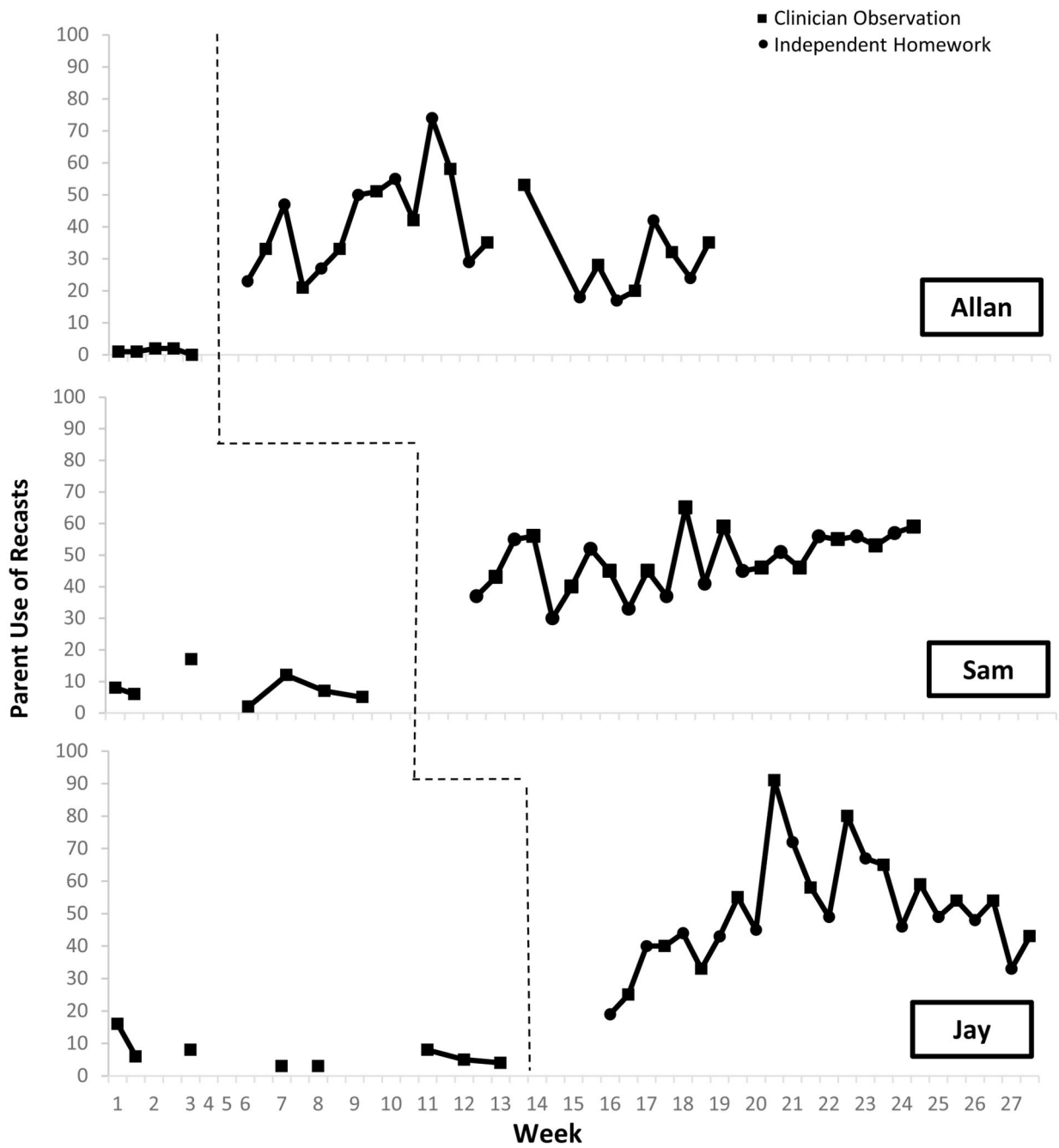
**Figure 2.**  
Frequency of Parent Strategy Use: WH-Questions

Author Manuscript

Author Manuscript

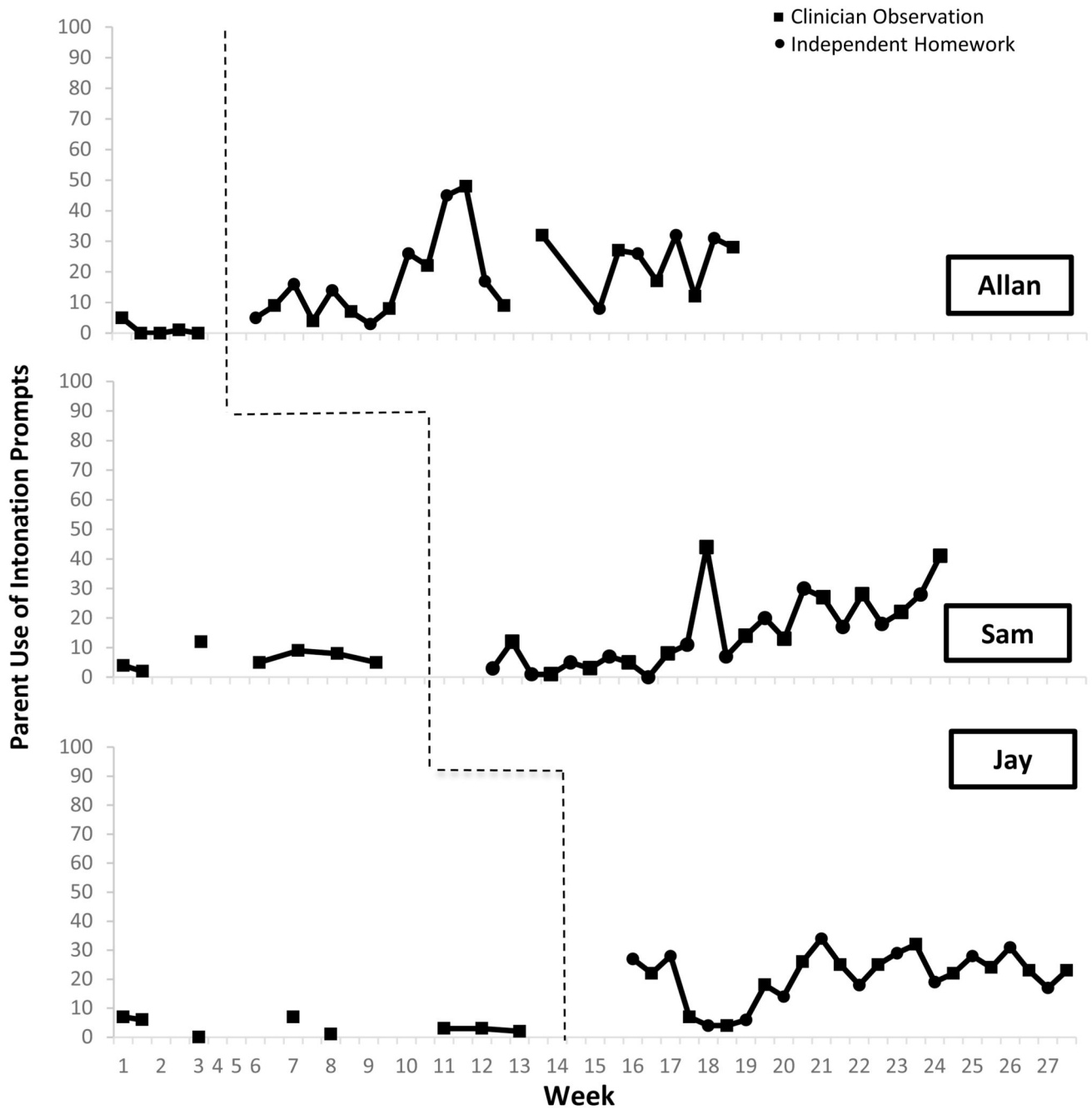
Author Manuscript

Author Manuscript



**Figure 3.**  
Frequency of Parent Strategy Use: Recasts





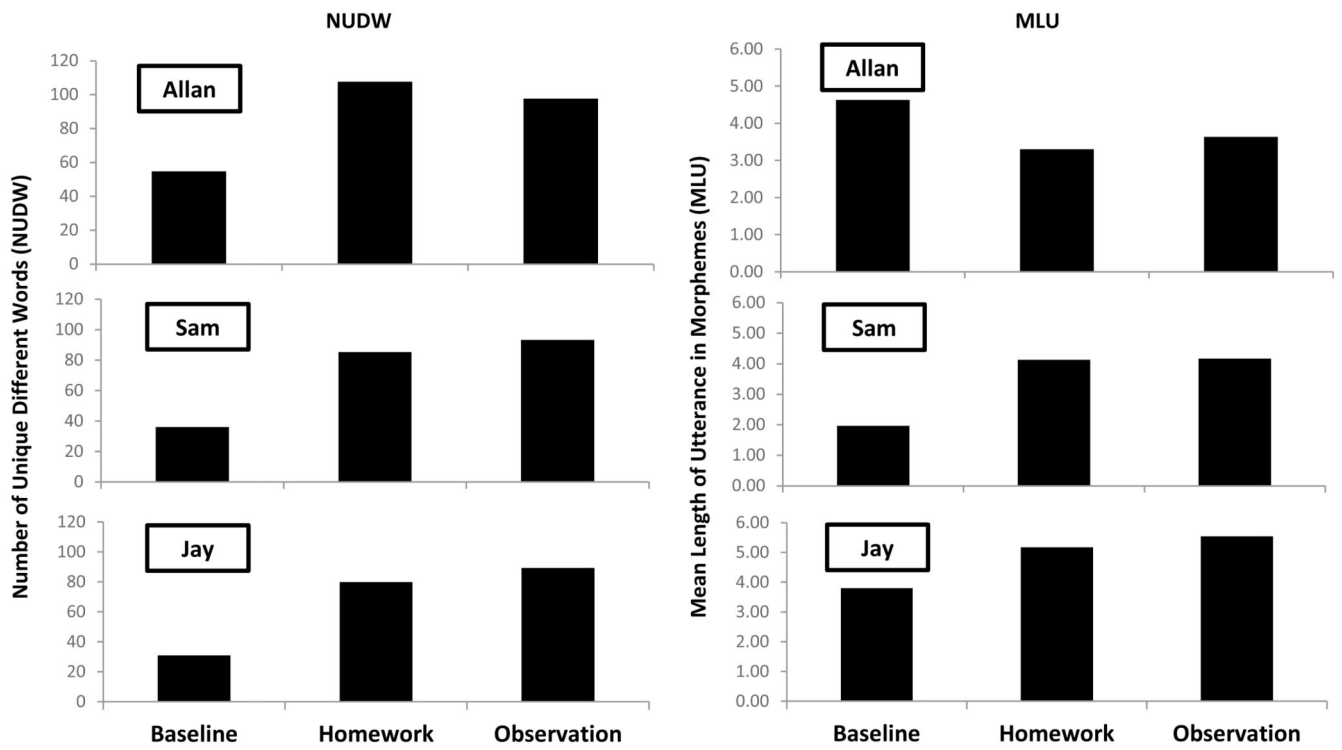
**Figure 4.**  
 Frequency of Parent Strategy Use: Intonation Prompts

Author Manuscript

Author Manuscript

Author Manuscript

Author Manuscript



**Figure 5.** Child number of unique different words (NUDW) and mean length of utterance in morphemes (MLU) produced during baseline, independent homework, and clinician observation sessions. The number of different words represents the average number of unique words produced during the shared reading of each different book that was utilized during the three session types.

**Table 1**  
**Descriptive Characteristics of Participating Dyads at Pre-Treatment Assessment Visit**

	Dyad 1	Dyad 2	Dyad 3
<u>Child Characteristics</u>			
Chronological Age	11-0	11-7	10-10
Nonverbal Cognitive Level <sup>1</sup>			
Age Equivalent	4-0	4-1	5-4
IQ	42	40	56
Receptive Vocabulary <sup>2</sup>			
Age Equivalent	5-11	6-4	7-4
Standard Score	61	63	76
Expressive Vocabulary <sup>3</sup>			
Age Equivalent	6-3	7-5	7-10
Standard Score	71	76	82
Expressive Syntax/Grammar <sup>4</sup>			
Age Equivalent	4-6	5-8	5-5
Standard Score	44	54	54
Autism Symptom Severity <sup>5</sup>	7	9	3
<u>Maternal Characteristics</u>			
CA	50	41	33
IQ <sup>6</sup>	103	103	102
Education (yrs)	Some College	Graduate Degree	

<sup>1</sup>Leiter International Performance Scales: Brief IQ (Miller & Roid, );

<sup>2</sup>Peabody Picture Vocabulary Test (Dunn & Dunn, );

<sup>3</sup>Expressive Vocabulary Test;

<sup>4</sup>Comprehensive Assessment of Spoken Language: Sentence Formulation;

<sup>5</sup>Autism Diagnostic Observation Schedule;

<sup>6</sup>Kaufmann Brief Intelligence Test.

**Table 2**  
**Illustrated Books Used During Baseline and Intervention Sessions**

Week	Allan	Pages	Sam	Pages	Jay	Pages
BL 1	The Perfect Pet	14	Mr. Gumpy's Motor Car	20	Duck on a Bike	16
BL 2	Cow Can't Sleep	21	Carl's Birthday	28	I Just Forgot	26
BL 3	Lost and Found	30	I Just Forgot	26	Harry the Dirty Dog	20
BL 4	Duck on a Bike	16	The Perfect Pet	14	If You Give a Dog a Donut	28
BL 5	I Took My Frog to the Library	15	Ready for Anything	25	Mr. Gumpy's Motor Car	20
BL 6			Pancakes for Breakfast	28	Ready for Anything!	25
BL 7			Harry the Dirty Dog	20	That's Good! That's Bad!	21
BL 8					Cow Can't Sleep	21
TX 1	Cow Can't Sleep	21	I Just Forgot	26	Duck on a Bike	16
TX 2	Duck on a Bike	16	Carl's Birthday	28	Harry the Dirty Dog	20
TX 3	Harry the Dirty Dog	20	Harry the Dirty Dog	20	Cow Can't Sleep	21
TX 4	Duck on a Bike	16	Carl's Birthday	28	Duck on a Bike	16
TX 5	If You Give a Dog a Donut	28	I Took My Frog to the Library	15	I Just Forgot	24
TX 6	I Just Forgot	26	Octopus Soup	27	If You Give a Dog a Donut	28
TX 7	I Took My Frog to the Library	15	Mr. Gumpy's Motor Car	20	Lost and Found	28
TX 8	I Just Forgot	26	I Took My Frog to the Library	15	If You Give a Dog a Donut	28
TX 9	Lost and Found	30	The Perfect Pet	14	Mr. Gumpy's Motor Car	20
TX 10	The Perfect Pet	14	Pancakes for Breakfast	28	Ready for Anything	25
TX 11	That's Good! That's Bad!	21	Ready for Anything	25	That's Good! That's Bad!	21
TX 12	The Perfect Pet	14	Pancakes for Breakfast	28	Ready for Anything	25

**Table 3**  
**Frequency and Percent Agreement for Clinician Coaching Behaviors**

Categories of Clinician Coaching	Child Participant		
	Allan	Sam	Jay
<u>General</u>			
Mean Frequency	35	48	33
Range	5-65	21-81	12-53
Percent Agreement	86%	95%	91%
<u>Wh-Questions</u>			
Mean Frequency	12	21	17
Range	3-19	12-32	7-32
Percent Agreement	95%	98%	97%
<u>Recasts</u>			
Mean Frequency	7	20	8
Range	0-19	4-44	2-20
Percent Agreement	86%	82%	88%
<u>Intonation Prompts</u>			
Mean Frequency	11	7	8
Range	0-25	0-20	3-13
Percent Agreement	95%	90%	97%
<u>Behavior Management</u>			
Mean Frequency	4	3	2
Range	0-14	0-7	0-7
Percent Agreement	90%	85%	89%
Mean Length of Coaching Sessions (mins:secs)	21:14	19:34	17:16

**Table 4**  
**Interobserver Agreement for Variables Coded from Baseline, Homework/Data Collection**  
**and Observation/Data Collection Sessions: Means (and ranges)**

Dyad	Dependent Variable	Experimental Session		
		Baseline	Homework	Clinician Observation
		<i>M%</i> (range)	<i>M%</i> (range)	<i>M%</i> (range)
1 <sup>a</sup>	Child Repetitions	91% (67-100)	79% (53-100)	75% (25-100)
	Child Non-Story Utterances	100%	76% (0-100)	91% (64-100)
	Child Mazes	80% (0-100)	95% (83-100)	97% (86-100)
	Child Story Related	100%	98% (92-100)	98% (96-100)
	Child Total Utterances	99% (97-100)	97% (92-100)	96% (92-99)
	Parent Response Models	100%	92% (86-100)	90% (82-98)
	Parent Use of Wh-Question	80% (0-100)	93% (82-100)	94% (75-100)
	Parent Recasts	90% (50-100)	88% (78-100)	87% (68-100)
	Parent Intonation Prompts	100%	99% (96-100)	99% (88-100)
2 <sup>b</sup>	Child Repetitions	86% (0-100)	92% (0-100)	79% (0-100)
	Child Non-Story Utterances	86% (0-100)	91% (67-100)	99% (91-100)
	Child Mazes	100%	79% (0-100)	88% (0-100)
	Child Story Related	99% (98-100)	99% (99-100)	100%
	Child Total Utterances	99% (98-100)	99% (98-100)	99% (98-100)
	Parent Response Models	93% (86-100)	87% (72-97)	89% (85-97)
	Parent Use of Wh-Question	91% (80-100)	92% (81-98)	93% (87-98)
	Parent Recasts	89% (80-100)	87% (65-96)	86% (79-93)
	Parent Intonation Prompts	100%	100%	100%
3 <sup>c</sup>	Child Repetitions	100%	96% (50-100)	83% (0-100)
	Child Non-Story Utterances	85% (0-100)	81% (0-100)	90% (50-100)
	Child Mazes	100%	83% (0-100)	100%
	Child Story Related	100%	100%	100%
	Child Total Utterances	99% (97-100)	99% (98-100)	99% (99-100)
	Parent Response Models	95% (80-100)	90% (79-100)	92% (81-100)
	Parent Use of Wh-Question	96% (67-100)	91% (72-100)	92% (83-100)
	Parent Recasts	91% (67-100)	82% (67-100)	91% (70-97)
	Parent Intonation Prompts	100%	99% (94-100)	99% (96-100)

<sup>a</sup>Dyad 1: 5 Baseline sessions, 11 Homework Sessions, 12 Clinician Observation Sessions

<sup>b</sup>Dyad 2: 7 Baseline sessions, 12 Homework Sessions, 12 Clinician Observation Sessions

<sup>c</sup>Dyad 3: 8 Baseline sessions, 12 Homework Sessions, 12 Clinician Observation Sessions

**Table 5**  
**Pre- and Post-Intervention Expressive Language Sampling: Examiner and Mother**

	Allan		Sam		Jay	
	Pre-TX	Post-TX	Pre-TX	Post-TX	Pre-TX	Post-TX
<u>Narration: Mother</u>						
Book	C	D	D	C	C	D
Duration (mins:secs)	7:35	19:53	8:21	19:36	4:42	14:05
NDW	137	158	82	203	45	139
MLU	3.50	2.55	2.43	3.20	2.73	3.85
Total Child Utterances	146	276	105	274	33	124
TNW	416	583	220	753	64	421
<u>Narration: Examiner</u>						
Book	A	B	B	A	A	B
Duration (mins:secs)	3:06	5:04	4:48	6:52	7:55	5:07
NDW	38	60	45	99	146	58
MLU	4.71	5.57	5.50	6.23	7.28	6.74
Total Child Utterances	31	47	21	66	75	23
TNW	136	243	82	357	512	136

Note: Book A = Frog Goes to Dinner; Book B = Frog on His Own; Book C = Frog, Where Are You; Book D = One Frog Too Many. Pre-TX = Pretreatment Assessment. Post-TX = Post-treatment Assessment.

Author Manuscript

Author Manuscript

Author Manuscript

Author Manuscript