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Publication Date

2018-03-01

DOI

10.1016/j.jcomdis.2018.02.007

Peer reviewed

HHS Public Access

Author manuscript

J Commun Disord. Author manuscript; available in PMC 2019 March 01.

Published in final edited form as:

J Commun Disord. 2018 ; 72: 64–76. doi:10.1016/j.jcomdis.2018.02.007.

Inferential language use by school-aged boys with fragile X syndrome: Effects of a parent-implemented spoken language intervention

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Abstract

This study examined the impact of a distance-delivered parent-implemented narrative language intervention on the use of inferential language during shared storytelling by school-aged boys with fragile X syndrome (FXS), an inherited neurodevelopmental disorder. Nineteen school-aged boys with FXS and their biological mothers participated. Dyads were randomly assigned to an intervention or a treatment-as-usual comparison group. Transcripts from all pre- and post-intervention sessions were coded for child use of prompted and spontaneous inferential language coded into various categories. Children in the intervention group used more utterances that contained inferential language than the comparison group at post-intervention. Furthermore, children in the intervention group used more prompted inferential language than the comparison group at post-intervention, but there were no differences between the groups in their spontaneous use of inferential language. Additionally, children in the intervention group demonstrated increases from pre- to post-intervention in their use of most categories of inferential language. This study provides initial support for the utility of a parent-implemented language intervention for increasing the use of inferential language by school aged boys with FXS, but also suggests the need for additional treatment to encourage spontaneous use.

Keywords

parent-implemented language intervention; intellectual disability; inferential language; telehealth

1. Introduction

School-aged males with fragile X syndrome (FXS) have difficulty using spoken language to engage in back-and-forth interactions with communication partners, even doing more poorly in this regard than younger typically developing (TD) children of the same cognitive levels

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(Abbeduto, Brady, & Kover, 2007). Unfortunately, there are no evidence-based language intervention approaches that have been designed and validated for older school-aged children or adolescents with this disorder. To address this concern, a parent-implemented, narrative-based language intervention was developed to target spoken language development in school-aged children with FXS, with preliminary evidence of efficacy (McDuffie et al., 2016, 2017). Although some interventions address narrative language development by teaching the elements of story grammar (e.g., Petersen, Gillam, & Gillam, 2008; Petersen, Gillam, Spencer, & Gillam, 2010), the intervention developed by McDuffie and colleagues targeted improvements in spoken language more generally by situating intervention activities within the context of shared storytelling activities using wordless picture books. Thus, the broad goal of this intervention approach is to support sustained verbal interactions between mothers and their children with FXS using wordless picture books, thereby creating the opportunity for learning and practicing new vocabulary and syntax. Numerous studies in other populations of children with language impairments have suggested that intervention activities that involve the sharing of wordless picture books with caregivers can support the development of these types of language skills (Hoffman, 2009; Kaderavek & Justice, 2002; Zevenbergen, Whitehurst, & Zevenbergen, 2003).

The narrative-based language intervention developed by McDuffie and colleagues was unique in two respects: (a) mothers were trained to be their child's partners during shared storytelling and (b) the intervention was delivered into the family home by means of distance video-teleconferencing (McDuffie et al., 2016, 2017). As a result of the intervention, mothers increased their use of targeted language facilitation strategies (i.e., models of story-related vocabulary and grammar, semantic and grammatical recasts, wh-questions, and intonation prompts; e.g., Desmarais, Nadeau, Trudeau, Filiatrault-Veilleux, & Maxès-Fournier, 2013; Dunst, Williams, Trivette, Simkus, & Hamby, 2012; van Kleeck, Vander Woude, & Hammett, 2006). There were also substantial increases in child vocabulary as measured by the number of different words participating children used at the post-treatment. Although the intervention was designed to lead to more sustained verbal interactions and enhanced vocabulary and syntax, previous research with other populations suggests that the adult language support strategies targeted in this intervention as well as the shared narrative context could also indirectly promote inferential language use (i.e., language that goes beyond describing concrete events depicted in story illustrations; van Kleeck et al., 2006) by the children who participated in the intervention.

Even before children learn to read, the ability to generate inferences in response to the information presented orally or visually in a storybook is an important component of narrative language competence (Kendeou, Bohn-Gettler, White, & van den Broek, 2008). As children go on to acquire the ability to decode text, inferencing provides a critical scaffold for reading comprehension (Tompkins, Guo, & Justice, 2013). Thus, the ability to use inferential language is an important aspect of spoken language competence, and the foundation for literacy, for children and adolescents with neurodevelopmental disorders, including those with FXS. Additionally, during primary data coding and analyses for this parent-implemented shared storytelling intervention (McDuffie et al., 2017), the use of inferential language, which is infrequent in the language of individuals with FXS (Simon, Keenan, Pennington, Taylor, & Hagerman, 2001), was noted with surprising frequency.

These observations led us to examine systematically and in greater detail inferential language use to determine whether participation in the narrative-based spoken language intervention also resulted in unplanned increases in the use of inferential language by school-aged boys with FXS.

1.1 Behavioral phenotype of FXS

FXS is the leading inherited cause of intellectual disability (ID; Crawford, Acuña, & Sherman, 2001; Hagerman, 2008). Because FXS is X-linked, males tend to be affected more than females, both in the prevalence of the disorder and in the severity of associated deficits. Males with FXS generally have IQ scores in the range of intellectual disability (< 70; Hessl et al., 2009), as well as other more specific cognitive impairments, including inattention and deficits in executive functioning (Cornish, Scerif, & Karmiloff-Smith, 2007; Loesch, Huggins, & Hagerman, 2004; Turk, 1998). In addition, males with FXS also frequently display symptoms of autism spectrum disorder (ASD), with as many as 60% of them having behaviors that are prevalent enough to meet diagnostic criteria for an ASD (Harris et al., 2008).

Deficits in acquiring and using spoken language to communicate effectively with others are ubiquitous among males with FXS. Although many boys with FXS demonstrate multiword syntax by adolescence (i.e., they can produce at least 3-word utterances on a daily basis), they are less able to participate in sustained interactions around a shared topic or use pragmatically appropriate language in social interactions (Abbeduto et al., 2007). Social anxiety, a lack of focused attention, and limited linguistic abilities may contribute to the presence of tangential, perseverative, and off-topic language (Keller-Bell & Abbeduto, 2007; Sudhalter & Belser, 2001; Roberts et al., 2007a), which are frequently observed during conversational interactions with boys with FXS. In fact, pragmatic skills in individuals with FXS are delayed compared to what would be expected based upon other language, cognitive, and social-emotional skills (Abbeduto & Hagerman, 1997). Males with FXS not only demonstrate pragmatic deficits, but also have co-occurring symptoms of ASD (Klusek, Martin, & Losh, 2013). Thus, boys with FXS are likely to have significant limitations in their ability to use and understand inferential language given that individuals with ASD also demonstrate deficits in pragmatics and in the use of inferential language in social communication (Bodner, Engelhardt, Minshew, & Williams, 2015; Dennis, Lazenby, & Lockyer, 2001). Boys with FXS also tend to use less complex grammatical forms than those used by their mental-age matched TD peers (Finestack & Abbeduto, 2010) while also demonstrating receptive and expressive vocabulary skills that are commensurate with those of their mental-age matched TD peers and as well as their nonverbal cognitive level (Abbeduto et al., 2007; Roberts et al., 2007b).

Additionally, several studies have examined the use of narrative language in FXS employing a variety of methods, including a study by Finestack, Palmer, & Abbeduto (2012) in which the Narrative Scoring Scheme (NSS; Heilmann, Miller, Nockerts, Dunaway, 2010) was used. Other studies of narrative language abilities in FXS have included more fine-grained, frequency-based approaches to examine the episodic structure of narratives (Channell, McDuffie, Bullard, & Abbeduto, 2015) as well as the use of inferential language to provide

explanations for, and evaluations of, events and character actions and reactions (Ashby, Channell, & Abbeduto, 2017). Collectively, these studies document delays relative to chronological age expectations on all measures for individuals with FXS. The delays, however, are generally consistent with developmental-level expectations. Moreover, in these studies, individuals with FXS outperformed those with Down syndrome on some measures, perhaps demonstrating a relative strength in some aspects of narrative language ability. Thus, the narrative context might be particularly useful for training new language skills in FXS.

Given the transactional nature of language learning (e.g., Dickinson & McCabe, 2001; McGinty, Justice, Zucker, Gosse, & Skibbe, 2012), reduced opportunities to engage in productive and sustained verbal interactions with others may, over time, cumulatively limit opportunities for language learning. These limited language-learning opportunities likely compound the cognitive impairments and further impede progress across many language domains over time, including expressive vocabulary, morphosyntax, and pragmatics (Abbeduto et al., 2007). Therefore, the role of the parent may be especially important in supporting language learning for children with FXS. In fact, research has shown that early and sustained verbal language input that is responsive to the child's focus of attention and ongoing communication attempts is positively related to language outcomes across childhood for children affected by FXS (Brady, Warren, Fleming, Keller & Sterling, 2014; Warren, Brady, Sterling, Fleming, & Marquis, 2010). A parent-implemented intervention can support the mother's acquisition of strategies that can, in turn, mediate the effect of the language intervention on the enrolled children.

1.2 Literal and inferential language

The types of spoken language that can be elicited during narrative-based intervention activities range along a continuum from literal to inferential (Westby, 1984). In the context of shared storytelling, child story-related utterances are literal to the extent that all of the information required for that response is overtly depicted in the illustration that the child is viewing. Thus, making inferences requires the speaker to fill in information that is not explicitly available in the pictures of a book (Kendeou et al., 2008). Inferential language allows children to understand and describe how individual pieces of a story contribute to a cohesive whole (Dougherty Stahl, 2004). The ability to generate inferences is also central to story comprehension regardless of whether the story is presented orally, in pictures, or in written text (Kendeou et al., 2008). Moreover, inferencing involves causally connecting events in the story, making predictions about the story, and interpreting events in the story according to one's world knowledge (Kintsch & Kintsch, 2005; van Kleeck, Gillam, Hamilton, & McGrath, 1997). Inferencing is especially important in the context of book sharing because interpreting the actions of characters requires the ability to understand that these actions are both goal-directed and causally related (Tompkins et al., 2013). Without the ability to use causal language, a story retell would stand as a collection of unrelated events. For example, in the book The Perfect Pet (Palatini & Whatley, 2003), there is a girl named Elizabeth who wants to find a pet. Inferential language produced by some of the children who read this story included, "She felt sad because she wanted a pet," and "Elizabeth is going to ask her parents for a horse." The first utterance conveys the character's mental state and an understanding of causal relationships, neither of which is directly expressed in the

picture, whereas the second is a prediction about what Elizabeth is going to do in the future to achieve her goal, and again, not directly expressed in the picture.

Beyond its contributions to spoken narrative language, numerous studies have shown that the use of inferencing is related to language comprehension in early childhood (Kendeou et al., 2008) and to reading comprehension for older elementary school-aged children (Cain, Oakhill, & Bryant, 2004; Hogan, Sittner Bridges, Justice, & Cain, 2011; Reese, Suggate, Long, & Schaughency, 2010; van Kleeck, 2008). Successful use and comprehension of inferential language is essential for both social and academic contexts and involves high level social-cognitive processes, including being able to understand the intentions (Happé, 1995) and mental states (Dennis et al., 2001) of others. Therefore, it is likely that deficits in inferential language also affect the ability of individuals with FXS to engage in sustained, socially-appropriate interactions during late childhood and adolescence.

Based on the work of Kendeou et al. (2008) and McGinnis et al. (2008), Tompkins et al. (2013) examined the ability of preschool-aged children to make inferences in a story generation task while viewing the wordless picture book, *Frog Goes to Dinner* (Mayer, 1974). The authors identified ten categories of inferential language which included: the action by which an agent's goal is achieved, character activities not related to a goal, the state of an object, the story setting, causal antecedents and consequences, character emotions, character dialogue, and a character's thoughts, perceptions, or physical characteristics. Utterances that can be assigned to these categories represent spoken language that goes beyond a simple description of the pictured events of the story.

Tompkins et al. (2013) found that typical preschoolers produced an average of 29 inferences while generating the story of *Frog Goes to Dinner*, and that the most frequent categories of inferential language were character activities that were unrelated to a goal (approximately 11 instances per story) and character state (approximately 5 instances per story). Children also produced utterances describing character emotions and character dialogue (approximately 3 and 2.5 instances per story, respectively). Descriptions of character goals and actions related to achieving those goals were infrequently mentioned as were causal antecedents and consequences. Their findings are relevant to the current study because the inference generation skills of boys with FXS are likely to be far closer to those of younger, mental-age matched TD children than age-matched peers. Thus, the coding scheme and results of Tompkins et al. (2013) was used as the framework for examining the inferences produced by the boys with FXS who participated in the current study.

1.3 The origins and development of inferential language competence

During the preschool years, children often participate in shared book reading with caregivers and these interactions expose children to rich and diverse forms of narrative language. The ability to make inferences is a skill that is gained through repeated exposures to shared book reading, as well as other experiences that do not simply refer to the here and now. At first, the adult partner in shared book reading scaffolds and elicits language by commenting on and asking questions about the literal aspects of the story depicted in the illustration (van Kleeck, 2008). As children get older, adult partners begin to talk about more sophisticated aspects of the story, including commenting on mental states and attitudes of the characters,

causal relationships between events, making predictions, and making connections between the story and general world knowledge. These shared storytelling opportunities also provide children with exposure to new vocabulary words that are reinforced through the story context (Desmarais et al., 2013; van Kleeck et al., 1997). With repeated exposure to shared stories, children begin to assume a greater role in telling the story by contributing more information themselves (van Kleeck et al., 2006).

A delayed trajectory, however, may be observed in book sharing between adults and children with language impairments. These children are less likely to engage in shared story-telling and to receive input that would encourage inference generation; that is, in response to child phenotypic characteristics, parents may provide fewer language models and ask fewer questions when engaged in book sharing than parents of children who are typically developing (e.g., Vander Woude, van Kleeck, & Vander Veen, 2009). Furthermore, adults seem less able to adjust their language input to the developmental level most optimal for children with language impairments (van Kleeck & Vander Woude, 2003).

Importantly, for children at risk for, or experiencing language impairments, the types of models and prompts that are used by adult partners during book sharing can scaffold the child's use of inferential language (Beck & Clarke-Stewart, 1998; Vander Woude & Koole, 2000; Zucker, Justice, Piasta, & Kaderavek, 2010). In a meta-analysis of adult book sharing strategies, Dunst and colleagues (2012) identified several adult strategies that best supported language and literacy outcomes for young children. These included the use of open-ended questions, providing or requesting decontextualized information about the story, relating events in the story to the child's personal experiences, and asking children to make predictions about different aspects of the story. Hogan et al. (2011) recommended the use of shared reading activities as a naturalistic and developmentally appropriate context within which adults can embed the use of explicit strategies to support children's language. Because the parent-implemented narrative language intervention examined in the current study targeted the use of open-ended questions, encouraged parents to model and prompt the use of more advanced vocabulary and grammar, and to provide developmentally advanced recasts of child responses, there was reason to expect that the use of these strategies would support inferential language use by the enrolled children. Thus, the current analysis examined whether boys enrolled in the intervention increased their overall frequency of use of inferential language during the shared storytelling activities with their mothers, whether there were differences between the intervention and comparison groups in the use of prompted versus spontaneous inferential language, and whether there were changes in the frequency of use of specific common categories of inferential language as a result of the intervention.

1.4 Research Questions

Many school-aged boys with FXS have multiword speech but face challenges engaging in sustained interactions around shared topics of conversation (Abbeduto et al., 2007). Because the ability to use inferential language is critical to narrative language competence and academic achievement and is learned through participation in shared storytelling, the goal of the current post-hoc analysis was to examine the effects of a parent-implemented language

intervention on the inferential language skills of school-aged boys with FXS. The following research questions were addressed:

1. Does participation in a parent-implemented spoken language intervention result in changes in the frequency of children's use of inferential language during shared story-telling interactions with the mother?
2. Does participation in a parent-implemented spoken language intervention result in changes in the frequency of children's use of prompted versus spontaneous inferential language during shared story-telling interactions with the mother?
3. Does participation in a parent-implemented spoken language intervention result in changes in the frequency of children's use of common categories of inferential language during shared story-telling interactions with the mother?

2. Method

2.1 Participants

The current study includes 19 mother-child dyads, described in McDuffie et al. (2017). The dyads were randomly assigned to either an intervention or a treatment-as-usual comparison group using a random numbers list. Child participants were eligible for the study if they had a confirmed diagnosis of full mutation FXS, age of 10 to 17 years, and maternal report that a) English was the primary language spoken in the home, b) the child was able to use at least three-word utterances on a daily basis, and c) there were no uncorrected sensory or motor impairments that would affect participation in the intervention. The study was approved by the Institutional Review Board at the University of California, Davis, and parents provided written informed consent prior to study participation.

No significant differences were found between the groups in measures of nonverbal IQ on the Leiter-R (Roid & Miller, 1997), $t(17) = -.006$, $p = .996$, $d = .002$; receptive vocabulary on the Peabody Picture Vocabulary Test – 4th Edition (PPVT-4; Dunn & Dunn, 2007), $t(17) = .178$, $p = .861$, $d = .22$; or expressive syntax on the Comprehensive Assessment of Spoken Language (CASL; Carrow-Woolfolk, 1999), $t(17) = -.587$, $p = .565$, $d = .27$, completed at the pre-intervention visit. However, a marginally significant difference was found between the groups in age, $t(17) = -1.986$, $p = .063$, $d = .93$, with the comparison group's mean age (M = 12.26, SD = 1.13) being lower than that of the intervention group (M = 13.92, SD = 2.26). Therefore, we controlled for age in all subsequent analyses that examined between-group differences in the use of inferential language. Moreover, mean non-verbal mental ages for the comparison and intervention groups were 4.67 years (SD = .61) and 4.71 years (SD = .90) respectively, indicating that it was appropriate to compare the inference generation skills of the boys in the current study to those in the sample of four- and five-year-olds in the study by Tompkins et al. (2013). In addition, although a full autism evaluation was not completed, three participants in the intervention group met criteria for ASD on the Autism Diagnostic Observation Scale – 2nd Edition (ADOS-2; Lord et al., 2012), whereas none met criteria in the comparison group; however, the two groups were not significantly different in terms of autism symptom severity on the ADOS-2, $t(17) = -.907$, $p = .377$, $d = .42$. Furthermore, according to parent report, all children in the study had received speech and

language therapy at some point in their lives (M = 10.11 years, range 7–14 years). All but two children in the comparison group and all but one child in the intervention group were receiving speech and language services while enrolled in the study. Additional information regarding participant screening and recruitment procedures as well as participant characteristics can be found in McDuffie et al., 2017.

2.2 Materials

2.2.1 Books and Scripts—Approximately 30 illustrated children's books were digitized and any accompanying text was removed from each book. Books were modified as needed so each was between 19 and 21 pages in length. For each intervention book, written scripts were created for the mothers which summarized the salient story events page by page and provided examples of relevant vocabulary and grammar; the scripts also included examples of questions that could be asked.

2.2.2 Video-teleconferencing equipment—Equipment loaned to each family included a MacBook Pro® laptop computer, an iPad®, and a Bluetooth headset. Each family's books were uploaded to the iPad® using the Apple iBooks™ application. The mother used the headset during coaching sessions so that the child would not hear the interventionist's comments and prompts. The clinician also used a MacBook Pro® laptop computer to initiate video calls with the family using the built-in iSight® web camera and Skype™ software. Ecamm™ Call Recorder Software was used with Skype™ to record coaching, feedback, and data collection sessions.

2.3 Design and structure of the intervention program

For dyads in the treatment group, the intervention consisted of: (a) at least one behavior support session administered by a Board-Certified Behavior Analyst (BCBA) that focused on antecedent- and consequence-based behavioral strategies (e.g., using a first/then card, implementing a token economy) to decrease or prevent challenging behaviors, reinforce appropriate behaviors, and increase engagement (occasionally, more than one behavior support session was deemed necessary by the clinical team in order to add additional strategies to the child's individualized behavior plan); (b) two parent education sessions focused on the rationale for and logistics of the intervention, and the language facilitation strategies; (c) weekly clinician coaching sessions delivered to the parent/child dyad via Skype™ during which the clinician provided the mother with real-time models, prompts, and reinforcement; (d) weekly homework sessions recorded by the parent using PhotoBooth® and uploaded electronically to the clinician using Dropbox™; (e) weekly clinician feedback sessions via Skype™ to discuss the homework session with the mother; and (f) weekly data collection sessions recorded by the clinician using Skype™ during which the mother and child retold that week's book for the final time, without any intervention from the clinician. Coaching, homework, feedback, and data collection sessions were completed each week during the 12-week intervention.

At the pre-intervention assessment clinic visit, the mother in each family randomized into the intervention group chose 12 books to use during the course of the intervention. One book was used during each week of the intervention for all sessions (i.e., coaching, homework,

feedback, and data collection). A different book was used in each week of the intervention by any given family. Prior to the coaching session each week, the parent and child selected which book they wanted to use for that week's intervention sessions. See Appendix A for a list of the books used in the intervention.

Dyads in the treatment-as-usual comparison group also received a behavior support session administered by a BCBA. Following this, they were sent two paperback books once a month during the 12 weeks of the intervention for a total of six books, but did not have any contact with intervention clinicians during this time. The books sent to the families in the comparison group were similar to the books that were used in the intervention, but they were not digitized or modified in any way. Mothers in the treatment-as-usual group were not provided with scripts for these books or any instructions on how or when to use them. See Appendix B for a list of the books sent to the families in the comparison group.

Additionally, all dyads completed three shared story-telling interactions that were recorded in the home via Skype™ during the two weeks prior to, and following, the 12-week intervention, and these served as the language samples for deriving the dependent measures for the current study. Three pairs of books were selected for these language samples and counterbalanced across participants and time points such that, for any given family, one book from each pair was used at the pre-intervention time point and the other was used at the post-intervention time point. The books in each pair had the same author and were similar in style and content. Unlike the intervention books, no scripts were provided for these books. See Appendix C for a list of the pairs of books used for the language samples recorded in the home at the pre- and post-intervention time points.

2.4 Implementation fidelity

Given that the intervention is still in the early stages of development, fidelity criteria (i.e., indicating parental mastery of the targeted content) have not yet been established. However, maternal response to coaching and completion of independent homework sessions provide an indirect measure of adherence to, and implementation of, the intervention as prescribed.

2.4.1 Maternal response to coaching—In order to investigate maternal implementation fidelity, both the proportion of clinician prompts that the mother responded to and maternal spontaneous use of targeted intervention strategies during coaching sessions were analyzed in the early, middle, and late stages of the intervention (i.e., weeks 1–4, weeks 5–8, and weeks 9–12; Banasik et al., 2017). Average maternal response to coaching for wh-questions was high (above 90%) during all stages of the intervention. However, maternal responses to coaching for expansions and intonation prompts was around 70–75% in the early stage and increasing to around 90% by the late stage of the intervention. Mothers also increased their spontaneous use of all strategies during coaching from the early to late stages of the intervention. From the early to the late stage of the intervention, average spontaneous use of wh-questions increased from about 20 to 25 per session, expansions increased from about 27 to 39 per session, and intonation prompts increased from about 13 to 15 per session. These numbers are meant to be purely descriptive given that fidelity criteria have not yet been established for this intervention. However, they seem to suggest

that mothers demonstrated greater independence of strategy use over the course of the intervention.

2.4.2 Completion of independent homework sessions—As another measure of maternal implementation fidelity, we examined the number of homework sessions completed by each dyad as well as the length of time of each homework session. Each dyad completed all twelve homework sessions and mean session length was 14 minutes (SD = 4.5, range = 6 – 29). In comparison, coaching sessions were longer than homework sessions with a mean length of 19 minutes (SD = 4.5, range = 9 – 33), which is to be expected given the amount of clinician modeling, prompting, and reinforcement that occurred during coaching sessions.

2.5 Transcription, coding, and inter-observer agreement

2.5.1 Transcription—Trained research assistants transcribed digitized video-recordings of the pre- and post-intervention sessions completed in the home using SALT (Systematic Analysis of Language Transcripts; Miller & Iglesias, 2008). SALT is a software program that allows for systematic transcription, coding, and analysis of transcripts prepared according to standard child language conventions. The research assistants transcribed the video-recordings according to the procedures described by Abbeduto, Benson, Short, and Dolish (1995). In this procedure, a primary transcriber completes a first draft of the transcript which is reviewed and edited by a second transcriber, and then finalized by the primary transcriber. This transcription process has been shown to yield average interrater reliability of 90% (Kover, McDuffie, Abbeduto, & Brown, 2012). For the current study, agreement between independently completed transcriptions was completed for about 10% of the transcripts using the procedures described above. Inter-transcriber agreement was computed for per utterance agreement on the identification of unintelligible words/segments, abandoned/interrupted utterances, mazes, number of words, word identity, and utterance segmentation. Overall mean agreement was 93.86% across those dimensions, with a range of 91.22 to 96.05% across transcripts.

2.5.2 Coding of inferential language—The inference coding scheme was adapted from Tompkins et al. (2013). Each child story-related utterance in the finalized SALT transcripts was coded for use of prompted or spontaneous inferential language in the following categories: 1) character goal-related actions, 2) character activities not related to a goal, 3) causal antecedents and consequences, 4) character dialogue, 5) descriptions of object state and story setting, and 6) character states including character thoughts, perceptions, desires, emotions, personality traits, physical characteristics, and role. An inference was considered prompted if it was preceded by maternal use of an open-ended wh-question or a fill-in-the-blank prompt and the child's response was related to the maternal prompt; other child inferences were coded as spontaneous. Of course, not every child utterance contained inferential language. For example, simply labeling a character or object is not considered inferential language. Moreover, the children sometimes misinterpreted a character's goal, action, mental state, or role, or had difficulty understanding or describing a causal relationship. Inaccurate interpretations of the story often contained language (e.g., cognitive or mental state verbs) that would typically signal an inference; therefore, the coders referred to the illustrations while coding to ensure that the child was accurately interpreting the story

details. Inaccurate inferences were not credited to the child. Definitions and examples of each inference category are presented in Table 1.

2.5.3 Inter-observer agreement—Three members of the research staff were trained through consensus coding to utilize the inference coding scheme adapted from Tompkins et al. (2013). Following this, two members of the research staff independently coded 57 transcripts each and another trained observer independently coded a randomly selected 20% of these transcripts. Two-way random intra-class correlation coefficients were above .950 for all inference categories except setting (.853).

2.6 Data analysis

All statistical analyses were performed using IBM SPSS version 24. For the first research question, a two-way mixed ANCOVA with Time (Pre/Post) as the repeated measure, Group (Treatment/Comparison) as the between-participants factor, and chronological age as the covariate was used to determine whether changes in the frequency of children's use of inferential language could be attributed to participation in the shared storytelling intervention. The dependent measure in these analyses was a composite variable derived by averaging across the three storytelling interactions completed in the home at the pre- and at the post-treatment. This variable included all spontaneous and prompted inferences produced by each participating child. Because the distribution of this variable showed significant right-skew, a square-root transformation was applied, after which the distribution closely approximated a normal distribution.

For the second research question, two-way mixed ANCOVAs with Time (Pre/Post) as the repeated measure, Group (Treatment/Comparison) as the between-participants factor, and chronological age as the covariate were used to determine whether changes in the frequency of (a) prompted and (b) spontaneous inferential language could be attributed to participation in the shared storytelling intervention. Again, each dependent measure was a composite variable derived by averaging across the three storytelling interactions at pre- and post-treatment. These variables were also square-root transformed to account for right skew. After the transformation, their distributions closely approximated a normal distribution. For this analysis, the alpha level was adjusted to 0.025 (0.05/2) to control for multiple comparisons.

For the third research question, which included only those children assigned to the treatment group, related-samples Wilcoxon signed rank tests were used to examine changes in use of common categories of inferential language collapsed across prompted and spontaneous inferences from pre- to post-treatment. Tompkins et al. (2013) found that inferences related to goals, causal relations, and character states were significantly correlated with story comprehension and predictive of story comprehension after controlling for child age and expressive vocabulary. Therefore, these categories (i.e., making inferences about character goal-related actions, causal antecedents/consequences, and character states) were considered higher-level inferencing and were examined separately from lower-level inferencing categories (i.e., making inferences about character activities, character dialogue, and setting). For each category of inferential language, the dependent measures in these analyses were composite variables derived by averaging across the three storytelling interactions at

the pre- and at the post-treatment. Wilcoxon signed rank tests were used for these analyses because this question was exploratory in nature and many of the variables were of low frequency and not normally distributed. For these analyses, the alpha level was adjusted to 0.0167 (.05/3) to control for multiple comparisons.

3. Results

3.1 Research question 1: Child use of inferential language

Table 2 displays untransformed means, standard deviations, and ranges for the frequency of child use of inferential language observed during the shared storytelling activities at the pre- and post-treatment. The two-way mixed ANCOVA on the transformed variable yielded a significant Time × Group interaction, $F(1,16) = 34.642$, $p < .001$, $\eta^2_{partial} = .684$. At the pre-treatment, boys in the intervention group and boys in the comparison group used inferential language at similar frequencies, whereas boys in the intervention used more inferential language at the post-treatment compared to the comparison group.

3.2 Research question 2: Child use of prompted and spontaneous inferential language

Table 2 also displays untransformed means, standard deviations, and ranges for child use of prompted and spontaneous inferential language observed during the shared storytelling activities at the pre- and post-treatment. For child use of prompted inferences, a two-way mixed ANCOVA on the transformed variable yielded a significant Time × Group interaction, $F(1,16) = 13.112$, $p < .001$, $\eta^2_{partial} = .594$. At the pre-treatment, boys in the intervention group and boys in the comparison group used prompted inferential language at similar frequencies, whereas boys in the intervention used more prompted inferential language at the post-treatment compared to the comparison group. For child use of spontaneous inferences, neither the main effects nor the interactions were significant.

3.3 Research question 3: Child use of common categories of inferential language

Table 3 displays means, standard deviations, and ranges for child use of common categories of inferential language at the pre-and post-treatment for children in the intervention group.

3.3.1 High-level inferencing—For character goal-related action inferences, a Wilcoxon signed-ranks test indicated that post-test ranks were significantly higher than pre-test ranks, $Z = -2.52$, $p = .012$. For causal antecedent/consequent inferences, a Wilcoxon signed-ranks test indicated that post-test ranks were not significantly higher than pre-test ranks, $Z = -2.20$, $p = .028$. For character state inferences, a Wilcoxon signed-ranks test indicated that post-test ranks were significantly higher than pre-test ranks, $Z = -2.60$, $p = .009$.

3.3.2 Low-level inferencing—For character activity inferences, Wilcoxon signed-ranks test indicated that post-test ranks were significantly higher than pre-test ranks, $Z = -2.70$, $p = .007$. For character dialogue inferences, a Wilcoxon signed-ranks test indicated that post-test ranks were not significantly higher than pre-test ranks, $Z = -2.04$, $p = .042$. For setting inferences, Wilcoxon signed-ranks test indicated that post-test ranks were significantly higher than pre-test ranks, $Z = -2.80$, $p = .005$.

4. Discussion

The current study provides preliminary evidence that participation in a distance-based parent-implemented language intervention, situated in the context of shared storytelling, can lead to increases in inferential language use for school-aged boys with FXS. These gains were observed despite the fact that inferential language was not explicitly targeted in the intervention. That is, mothers were not explicitly provided with information about the differences between literal and inferential language or prompted to target this type of utterance. Participants in both the intervention and treatment-as-usual comparison groups used minimal levels of prompted and spontaneous inferential language during pre-intervention distance sessions. This finding suggests that either the participants were not able to understand or interpret story elements beyond a literal level or that they were not being provided with sufficient opportunities to produce such language. However, participants in the treatment group demonstrated marked increases in their use of prompted, but not spontaneous, inferential language during the post-intervention sessions. This finding suggests that maternal prompting was driving the effect of overall increased use of inferential language in the intervention group, given that the groups did not differ in their use of spontaneous inferential language during the post-intervention sessions. That is, increases in maternal use of open-ended wh-questions and intonation prompts encouraged the children in the treatment group to attend to and make sense of less salient aspects of the story, which led to increases in their inference generation during the post-intervention sessions.

The fact that all mothers in the treatment group demonstrated increases in the targeted language support strategies (i.e., contingent semantic and grammatical recasts, wh-questions, and intonation prompts; McDuffie et al., 2017) and that there were increases in their child's use of inferential language is consistent with previous research on younger TD children. In past studies, these types of prompting strategies resulted in children using more inferential language (e.g., van Kleeck et al., 2006; Desmarais et al., 2013). For example, questions incorporating "why" and "how" can be used to prompt the use of child responses containing inferential language, even though the child might not initially use such language spontaneously when telling a story. Similarly, a mother could use an intonation prompt to elicit inferential language that a child might not use independently. For example, when telling the story Pancakes for Breakfast (dePaola, 1978) for the first time, a mother might say, "The lady had no maple syrup. She felt so *disappointed*," or "The cat thought the cream tasted *delicious*". During subsequent retellings, the mother could prompt the use of these previously introduced vocabulary words by using either a wh-question ("How did the lady feel when she discovered she was out of maple syrup?") or an intonation prompt ("The cat thought the cream tasted _____!"). Both examples would prompt the child to use inferential language as the former queries the lady's emotional state and the latter queries the way that the cream tasted to the cat. These descriptions go beyond the literal information conveyed in the illustrations of the book.

Additionally, participants in the treatment group engaged in more inferencing across most categories during post-intervention sessions compared to pre-intervention sessions, with particularly large gains in the participants' use of both character state and character activity inferences. These findings are similar to those found in Tompkins et al. (2013) in that

inferences relating to character state and character actions not related to a goal were the inference types most frequently produced by the TD preschoolers in their sample, suggesting that school-aged boys with FXS can generate the same types of inferences as TD children of a similar mental age. This finding may be due in part to the types of questions and intonation prompts that parents were coached to use during the 12-week intervention. Although the scripts were not written specifically to emphasize non-literal aspects of the stories, the clinician did sometimes suggest that the parents highlight and comment on character mental states, including cognitive and perceptual processes, as well as desires and emotions. For example, as mothers modeled mental state vocabulary and encouraged the child to attend to less salient aspects of the story or illustrations (e.g., facial expressions of characters), perhaps the participant was better able to generate inferences about character states. Marked gains were also observed for child use of character activity inferences. Past research has also shown that TD children can identify character activities and character states even before they enter kindergarten (Tompkins et al., 2013). Furthermore, the youth with FXS in the intervention group produced relatively low numbers of utterances containing inferences related to character dialogue, causal antecedents and consequences, setting, and goal-related actions (i.e., attempts), similar to the findings of Tompkins and colleagues (2013).

4.1 Limitations

One limitation of the study is that participants in the treatment group had at least three weekly practices with each book (i.e., during coaching, homework, and data collection sessions) during the 12-week intervention, but they had only one opportunity to see and talk about each novel book during the pre- and post-intervention sessions. It is possible that additional exposures to these books would have allowed them to produce more inferential language spontaneously or that mothers might begin to fade their use of prompts that targeted inferential language. Of course, an increase in spontaneous inferential language use could potentially be observed in both the treatment and comparison groups given multiple exposures to the books. However, it remains unlikely that the children in the comparison group would produce more prompted inferential language with additional exposures to the books considering that their mothers were not familiar with the targeted language support strategies that may have facilitated increased use of prompted inferences in the children in the intervention group.

Another component of the intervention program was the use of individualized scripts to accompany each picture book used during the intervention. The scripts were provided to familiarize the mothers with the content of the stories conveyed by the wordless picture books and to provide examples of the vocabulary and grammar that could be used to tell each story. These scripts, however, were not provided for the pre- and post-intervention sessions. Intervention effects on inferential language might have been more substantial had the scripts been tailored to specifically emphasize the use of inferential language and had they included questions involving prediction and background knowledge (e.g., van Kleeck et al., 2006). However, there was some inclusion of inferential language in the scripts as the mothers were modeling complex sentences (e.g., with linguistic and mental state verbs). Perhaps mothers in the intervention group who had exposure to these scripts during the 12-

week intervention were better able to notice, talk about, and prompt discussion about aspects of the story that required inferencing compared to the mothers in the comparison group.

Furthermore, given that this intervention is in the early stages of development, the treatment-as-usual comparison group did not receive any type of complimentary intervention from the study clinicians. Therefore, there were many differences in the experiences between the treatment and comparison groups that could have contributed to the languages differences observed between these groups. For example, although parents in both the intervention and comparison groups received a behavior support session with a BCBA, the intervention group had significantly more practice with, and exposure to, the behavior strategies, which could have influenced their performance in the post-intervention sessions by allowing them to better engage with the book. Additionally, having significantly more exposure to the technology used to implement the intervention and record the language samples in the home could have benefitted the intervention group. It is possible that the comparison group's performance in the post-treatment sessions was due at least in part to being less comfortable with distance video-teleconferencing. However, anecdotally, most participants in both the intervention and comparison groups had developed good rapport with the clinicians and study staff who were conducting the pre- and post-intervention calls and enjoyed seeing and interacting with them over Skype. Nevertheless, future iterations of the intervention should attempt to control for additional exposure to behavioral support strategies and technology, as well as comfort and familiarity with study staff, as confounding variables by scheduling frequent non-intervention-related calls with dyads in the non-treatment comparison group.

In addition, the comparison group was simply mailed books during the course of the intervention without ensuring that mothers engaged the child with the books for the same amount of time as the treatment group. Therefore, it is possible that simply having more exposure to shared storytelling activities led to increased use of inferential language by the children in the intervention group; that is, it is possible that the specific strategies the mothers were taught might not have been the active ingredient of change but rather simply participating in shared storytelling. However, a single-case, multiple-baseline study of the intervention by McDuffie et al. (2016) suggests that this is likely not the case. Before beginning the intervention, mothers in the single-case study had between five and nine weeks of shared storytelling experiences with their child in the form of baseline sessions and they demonstrated minimal use of the targeted maternal strategies. Moreover, these children showed little to no use of inferential language during these baseline sessions. However, upon the introduction of the intervention, increases in maternal strategy use as well as increases in inference generation were observed across all three dyads (Nelson et al., 2015). Therefore, it seems more likely that exposure to the mother's use of targeted strategies as opposed to exposure to the shared reading activities in general contributed to increases in inferential language use.

Another limitation of the present study is that there was significant variation in strategy acquisition and use across mothers in the treatment group and thus, variation in the dose of the intervention that the children received throughout the intervention. Indeed, because the intervention is in the early stages of development, fidelity criteria for maternal mastery have not yet been established. Therefore, the dose of the intervention that the children received

was highly variable and not tightly controlled. Although we observed changes in maternal spontaneous use of the targeted strategies over the course of the intervention, we do not yet know if that change was sufficient to benefit the children. In addition, although many mothers reported that they were using the language facilitation strategies outside of the intervention sessions during every day conversational interactions, we cannot be sure how often and to what extent children were exposed to the strategies outside of the coaching, homework, and data collection sessions. Future studies should carefully examine maternal implementation of the strategies and the effects of these strategies on child language, including inference generation.

4.2 Future directions

In future iterations of this intervention, it would be important to determine the effects of adding more inferential language to the scripts and providing explicit training during parent education sessions about the differences between literal and inferential language. This might affect the mothers' use of inferential language, and the changes in maternal modeling of inferential language might lead to increased use of inferential language by their children. Such more explicit targeting of inferential language might be needed to encourage spontaneous use by the children with FXS.

Inference use was observed to be highly variable at pre- and post-intervention in both groups. We hypothesize that variability could be due to multiple factors, including the use of books that varied in content as well as variable amount of time spent discussing a book during the sessions. Although naturalistic interventions such as this one could be expected to yield high levels of variability given that the number and type of parent prompts was not tightly controlled, it would be important to more fully investigate sources of variability in order to maximize the effectiveness of the intervention sessions. Additionally, the sample size in this study was too small to determine whether certain child or parent baseline variables predicted who responded best to treatment, yet this is an important question to address in future iterations of the intervention that include a larger sample of participants.

Furthermore, it would be interesting to determine whether the children's comprehension of inferential questions improved during and after the intervention. Based upon past research with young children, we would expect to see an increase in comprehension of questions containing inferential language along with increases in the production of inferential language (e.g., Desmarais, et al., 2013; Kendeou, et al., 2008; Tompkins et al., 2013; van Kleeck et al., 2006). It would also be interesting to see if children in the intervention group demonstrated increases in their overall use of inferential language over the course of the intervention week (i.e., from coaching to data collection) and between weeks or if there were changes in the ratio of use of prompted versus spontaneous inferential language. Another important future direction includes determining whether participation in the intervention leads to gains in inferential language during a naturalistic conversational interaction with the mother or an examiner. Moreover, this study only included males with FXS. Although many females with FXS are less likely than males to meet the study criteria for nonverbal IQ (<70), it will be interesting to examine differences between males and females in inferential language use in future studies of the intervention that include female participants.

In conclusion, this study indicates that a naturalistic parent-implemented narrative language intervention can increase the use of inferential language by school-aged children with FXS. To date, there are no known evidence-based narrative language interventions for school-aged or adolescent individuals with FXS. The framework of this intervention could easily be utilized for other groups of children with neurodevelopmental disorders and co-occurring language delays. Furthermore, language interventions for school-aged children and adolescents do not typically include the parent as the delivery agent for the intervention, yet this study shows that parents can be trained to use language facilitation strategies and implement them independently with school-aged children and adolescents. These mothers also reported to the clinician and SLP that they were generalizing the use of the language strategies to everyday conversations, suggesting that the role of parents as interventionists could have important implications by increasing the frequency and duration of child participation in sustained conversational interactions with their caregivers outside of the intervention context (McDuffie et al., 2016, 2017). In this way, the child could be exposed to more opportunities to hear and acquire more advanced vocabulary and syntax during every day routines and interactions. Additionally, this study demonstrates that shared storytelling using wordless picture books can be successfully utilized for older children with cognitive impairments. The intervention could easily be modified to be delivered in person in a classroom or in therapy sessions by a child's teacher or SLP and provides a viable option for an intervention format for school-aged and adolescent individuals with significant language delays. Future iterations of this intervention could be delivered, perhaps with modifications, to children with other neurodevelopmental disorders, such as those with Down syndrome and ASD. Finally, given the role of inferencing in both narrative comprehension (Kendeou et al., 2008) and reading comprehension (Cain et al., 2004; Hogan et al., 2011; Reese et al., 2010; van Kleeck, 2008), increasing inferential language competence could lead to more positive functional and academic outcomes for individuals with FXS and those with other disabilities.

Acknowledgments

This work was supported the UC Davis MIND Institute Intellectual and Developmental Disabilities Research Center funded by the Eunice Kennedy Shriver National Institute for Child Health and Human Development (U54 HD079125, PI: Leonard Abbeduto). We extend our deepest appreciation to the families who participated in this research. Preliminary results from this project were presented at the 50th Annual Gatlinburg Conference on Theory and Research in Developmental Disabilities (March 2017; San Antonio, TX).

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Appendix A. Intervention Books

Book	Author and Illustrator
Badger's Fancy Meal	Keiko Kasza
Carl's Birthday	Alexandra Day
Chalk	Bill Thomson
Charlie the Ranch Dog	Ree Drummond and Diane deGroat
Cow Can't Sleep	Ken Baker and Steve Gray
Duck on a Bike	David Shannon
Harry the Dirty Dog	Gene Zion and Margaret Bloy Graham
I Just Forgot	Mercer Mayer
I Took My Frog to the Library	Eric A. Kimmel and Blanche Sims
If You Give a Dog a Donut	Laura Numeroff and Felicia Bond
Lost and Found	Oliver Jeffers
Marley: Messy Dog	John Grogan and Richard Cowdrey
Mooncake	Frank Asch
Mr. Gumpy's Motor Car	John Burningham
Octopus Soup	Mercer Mayer
Pancakes for Breakfast	Tomie dePaola
Pete the Cat: Pete at the Beach	James Dean
Ready for Anything!	Keiko Kasza
That's Good! That's Bad!	Margery Cuyler
The Invisible Boy	Trady Ludwig and Patrice Barton
The Perfect Pet	Margie Palatini and Bruce Whatley

Appendix B. Books sets sent to dyads in the treatment-as-usual comparison group

Set	Book	Author and Illustrator
1	I Was So Mad	Mercer Mayer
	Secret Pizza Party	Adam Rubin and Daniel Salmieri
2	If You Give a Mouse a Cookie	Laura Numeroff and Felicia Bond
	Too Many Toys	David Shannon
3	Mr. Gumpy's Outing	John Burningham
	Those Darn Squirrels	Adam Rubin and Daniel Salmieri

Appendix C. Book pairs used for the mother-child language samples recorded in the home

Pair	Book	Author and Illustrator
A	Suddenly	Colin McNaughton

Pair	Book	Author and Illustrator
	Oops!	
B	If You Give a Pig a Party	Laura Numeroff and Felicia Bond
	If You Take a Mouse to School	
C	Just a Day at the Pond	Mercer Mayer
	Just a Little Music	

Table 1

Definitions and Transcribed Examples of Inferential Language in Each Category

Inference Type	Definition	Example
Character goal-related action	Reference to an action by which an agent's goal is achieved	Child: The wolf is gonna eat the pig. Child: He kicked the baby frog off the raft.
Causal antecedents/consequences	A connection between the current event and the previous context/event or an event that follows a current event	Parent: Why is the dog licking the boy's hand? Child: So he won't feel bad.
Character state	Reference to a character's thoughts	Child: He got an idea to cook something else.
	Reference to a character's emotions	Child: She's mad at the boy.
	What a character perceives	Child: He is watching the lady cook.
	What a character desires	Child: He doesn't want to get made into soup.
	Character personality trait or physical trait	Child: The dog was too loud. Child: He was very tall.
	Character role	Child: The store owner is reading.
Character activity	A character action that is not related to a goal	Parent: What are dad and sister doing? Child: Swimming.
Character dialogue	What a character says, or reference to character speech	Parent: What do you think his dad is telling him? Child: "Don't go too far."
Object state/Setting	A state of an object	Child: The guitar is broken.
	Setting of story event	Parent: Where are the crocodile and the boy now? Child: His bedroom.

Table 2

Child Frequency of Use of Total, Prompted, and Spontaneous Inferential Language During Language Samples in the Home (N=19)

	Pre-Treatment		Post-Treatment	
	Treatment Group	**Comparison Group**	**Treatment Group**	**Comparison Group**
	Mean (*SD*) Range	**Mean (*SD*) Range**	**Mean (*SD*) Range**	**Mean (*SD*) Range**
Total	11.53 (9.40) 0.75 – 29.00	13.36 (9.89) 0.75 – 33.75	28.35 (18.36) * 5.34 – 63.00	10.97 (10.38) 0.33 – 36.33
Prompted	6.38 (5.22) 0.25 – 16.00	8.42 (5.84) 0 – 15.25	23.10 (13.83) * 4.67 – 46.67	6.67 (5.73) 0.33 – 19.00
Spontaneous	5.15 (7.64) 0.50 – 25.50	4.94 (5.37) 0.75 – 18.50	5.25 (5.45) 0.67 – 16.33	4.30 (5.17) 0 – 17.33

Note.

* $p < .001$.

Table 3

Frequency of Use of Common Categories of Inferential Language in the Intervention Group During Language Samples in the Home (N=10)

	Pre-Treatment	Post-Treatment
Inference Category	**Mean (*SD*) Range**	**Mean (*SD*) Range**
Character Goal-Related Action	0.45 (0.50) 0 – 1.25	1.13 (0.97)* 0 – 3.00
Causal Antecedent/Consequence	0.18 (0.26) 0 – 0.75	1.23 (1.89) 0 – 6.00
Character State	4.20 (3.29) 0 – 10.25	7.55 (5.87)* 1.00 – 20.00
Character Activity	3.90 (3.99) 0.25 – 11.00	9.73 (6.58)* 1.67 – 19.00
Character Dialogue	0.78 (0.69) 0 – 2.25	2.33 (2.38) 0 – 6.67
Object State/Setting	2.03 (1.42) 0 – 4.00	6.37 (2.83)* 1.33 – 10.00

Note.

*$p < .0167$ (adjusted p-level for multiple comparisons).