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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, 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.

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 (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), 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

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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