



Research Paper

Play with objects in young males with fragile X syndrome: A preliminary study



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ABSTRACT

Using the Developmental Play Assessment, this preliminary study described the categories and levels of play with objects produced by 10 young boys with diagnoses of full mutation fragile X syndrome, the leading inherited cause of intellectual disability. Additionally, the study examined concurrent associations between child characteristics and three different summary level variables representing object play skills. Presentation Combinations (i.e., recreating structured configurations of objects) was the highest play level emerging or mastered for all participants. The number of toys touched during the play sample, an index of object interest, was positively related to standardized measures of receptive and expressive language while the number of different actions produced, an index of play diversity, was negatively related to autism symptom severity. Both variables were significantly related to the number of nonverbal communication acts children produced while interacting with their mothers in play. Clinical implications and future directions are discussed.

Learning outcomes: Readers will be able to: (1) define a framework for categorizing developmental levels of play; (2) discuss the constructs represented by three different summary level metrics of play with objects; (3) describe the relationship between object-play skills and child characteristics for young males with FXS.

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1. Introduction

Play with objects makes a critical contribution to many important developmental achievements during early childhood. Exploratory object play scaffolds problem solving skills and cognitive development as children learn about the properties of objects (Fenson & Schell, 1985; Nicolopoulou, 1993; Sutton-Smith, 1997), functional object play provides a social learning mechanism for acquiring the conventional uses of familiar objects (Ungerer, Zelazo, Kearsley, & O'Leary, 1981), and symbolic play with either replica or natural objects supports the development of representational thought and symbol use (Lillard, Pinkham, & Smith, 2011; Tomasello, Striano, Rochat, 1999). Importantly, by supporting engagement with other people, object play scaffolds developments in social interaction (Pierce-Jordan & Lifter, 2005), social communication, and language (McCune, 1995; Shore, O'Connell, & Bates, 1984; Tamis-LeMonda & Bornstein, 1990; Tamis-LeMonda & Bornstein, 1994). Although the emergence, developmental correlates, and predictive functions of play with objects have been widely studied

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in children with typical and atypical development, there are no published studies examining play with objects in children with fragile X syndrome (FXS), the leading inherited cause of intellectual disability (Crawford, Acuna, & Sherman, 2001). The phenotypic characteristics of young males with FXS, however, are likely to significantly affect both the developmental emergence of play skills and the amount of time spent productively engaged with objects. Thus, examining patterns and correlates of play with objects in young children with the FXS full mutation can further our understanding of the behavioral phenotype of this disorder. In the current study, we were interested in providing a descriptive analysis of the object-focused play skills of a group of young boys with FXS who were at the earliest stages of language development. We also examined concurrent associations between several different metrics of play with standardized and naturally occurring measures of cognition, communication, and language.

1.1. Fragile X syndrome

FXS is caused by a mutation in the *FMR1* gene which is on the X chromosome (Verkerk et al., 1991). The normal version of the *FMR1* gene is comprised of 5–54 CGG repeats, but individuals with FXS have expansions exceeding 200 repeats, which is termed the full mutation. The FXS full mutation leads to hypermethylation and transcriptional silencing of the gene such that its protein product, termed the fragile X mental retardation protein (FMRP), is reduced or absent (Oostra & Willemson, 2009). FMRP regulates protein synthesis at the synapse; thus, FMRP is important for cognition as it guides neuronal development and experience-dependent learning (Bassall & Warren, 2008). Prevalence estimates for the *FMR1* full mutation are 1 in 3500 males and 1 in 6000–8000 females (Coffee et al., 2009). Relative to males, females with the full mutation tend to be less affected, given the protective presence of an unaffected X chromosome.

1.2. Males with fragile X syndrome

Virtually all males with the *FMR1* full mutation have cognitive delays, with 85% having IQs between 40 and 55 (Hessl et al., 2009). Additionally, language delays are often more severe than would be expected based upon nonverbal cognitive level (Abbeduto, Brady, & Kover, 2007) due to other phenotypic characteristics that negatively impact the ways in which males with FXS are able to use spoken language during conversational interactions. For instance, the majority of males with FXS display hyperarousal and attentional difficulties (Cornish, Scerif, & Karmiloff-Smith, 2007; Scerif, Longhi, Cole, Karmiloff-Smith, & Cornish, 2012); anxiety and social withdrawal (Cordeiro, Ballinger, Hagerman, & Hessl, 2011; Kau, Reider, Payne, Meyer, & Freund, 2000); repetitive and stereotyped behaviors (Hall, Lightbody, Hirt, Rezvani, & Reiss, 2010); and other challenging behaviors, including aggression and self-injury (Hall, Lightbody, & Reiss, 2008; Symons, Clark, Hatton, Skinner, & Bailey, 2003). Challenging behaviors emerge early in toddlerhood for males with FXS (Symons et al., 2003; Symons, Byiers, Raspa, Bishop, & Bailey, 2010) and most frequently function to escape task demands or to obtain preferred items (Symons et al., 2003; Citation removed for blind review, 2014).

As many as 90% of males with FXS also display behaviors that are consistent with symptoms of autism, such as hand flapping, repetitive speech, and gaze aversion (Merenstein et al., 1996). Across studies, symptoms of autism are frequent and severe enough that approximately 60% of males with FXS receive a comorbid diagnosis based upon meeting gold-standard diagnostic criteria for an autism spectrum disorder (ASD) (Bailey, Mesibov, Hatton, Clark, Roberts, Mayhew, 1998; Demark, Feldman, Holden, & McLean, 2003; Hall et al., 2010; Harris et al., 2008). On average, boys with comorbid FXS and ASD have lower levels of nonverbal cognition (Hernandez et al., 2009; Citation removed for blind review, 2014; Wolff et al., 2012) and higher rates of challenging behaviors (Smith, Barker, Seltzer, Abbeduto, & Greenberg, 2012) than boys diagnosed with nonsyndromic ASD (i.e., ASD for which a genetic etiology has been ruled out).

1.3. Play in typically developing children and those with developmental disabilities

For typically developing children, play with objects emerges during infancy and, broadly speaking, progresses sequentially from the indiscriminate manipulation of objects to enacting experiences and events with symbolic actions. Thus, it is generally accepted that the emergence of symbols is concurrently reflected in children's actions during play as well as in their accomplishments in referential communication (Bloom, Tinker, & Scholnick, 2001; Ungerer & Sigman, 1984; McCune, 1995). Children with developmental disabilities are likely to produce fewer actions with objects, use less diverse and complex play, and be interested in fewer different objects than typically developing children at the same developmental levels (Lifter, Foster Sanda, Arzamarzki, Briesch, & McClure, 2011; Thiemann-Bourque, Brady, & Fleming (2012)).

Numerous researchers have proposed a variety of taxonomies that specifically describe the sequential stages of play through which children progress (Belsky & Most, 1981; Fenson, Kagan, Kearsley, & Zelazo, 1976; Lifter & Bloom, 1989; Lifter, Edwards, Avery, Anderson, Sulzer-Azaroff, 1988; McCune, 1995; Ungerer & Sigman, 1981). In the current study, we utilized the Developmental Play Assessment (DPA) developed by Lifter and colleagues (Lifter, 2000). The DPA was designed to assess the play activities of children with developmental disabilities; thus, it includes numerous qualitative subcategories that can be used to provide a more nuanced description of play in children who have less well-developed play skills (Lifter, 2000).

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