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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, & Fruend, 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; 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).

The play categories included in the current study represent four developmental levels of play with objects: Indiscriminate Play Actions, Discriminative Play Actions, Combinatorial Play Actions, and Pretend Play Actions. In typical development, indiscriminate play emerges at a developmental level of 4–6-months as the child produces simple actions involving visual exploration and tactile manipulation of objects (i.e., mouthing, shaking, banging; Vondra & Belsky, 1989; Williams, 2003). Discriminative play actions emerge at a developmental level of approximately 7–9-months, as the child begins to produce actions that are specific to the physical attributes of the manipulated objects (e.g., rolling a ball, turning a dial on a toy phone, pushing a vehicle with wheels). During this same developmental period, the child is able to separate objects from the configurations in which these objects are initially presented (e.g., removing pieces from a puzzle). Many researchers have suggested that the production of discriminative play actions supports development in cognition and problem solving.

By approximately 9-months of age, children begin to display combinatorial play actions as they bring together objects in ways that are constrained by the physical attributes of the objects (e.g., nesting cups inside one another, putting a peg in a hole; Belsky & Most, 1981). At a developmental level of 12-months, the child is able to combine two or more unrelated objects (e.g., putting objects into a container), and later combine objects based upon their specific physical attributes (e.g., stacking blocks, stringing beads; Fenson et al., 1976). The ability to simultaneously consider two or more objects enables children to explore, through active manipulation and visual inspection, the functional, spatial, causal, and categorical relationships between objects (Fenson & Schell, 1985). All of these types of play may be considered functional as they use objects as they were intended.

At a developmental level of approximately 13-months, children first begin to use objects to reproduce actions they have encountered during daily routines and they later begin to direct these conventional actions to play partners and inanimate figures (e.g., putting a spoon to mouth; Fenson & Schell, 1985). These types of actions demonstrate knowledge of the action that is conventionally associated with an object (Bigelow, MacLean, & Proctor, 2004). Fenson and colleagues (1976) noted that approximately one-half of typically developing 13-month olds and all of typically developing 20-month olds performed at least one functional play act when observed for 20 min with a variety of toys. Thus, at a cognitive developmental level between 13- and 20-months, we can anticipate that children with developmental disabilities will begin to produce play actions that include functional play with conventional objects.

Pretend play, the final play level designated by Lifter (2000) and considered in the current study, includes two categories of play with objects that are generally considered to be symbolic. The first category involves object substitutions in which a familiar action is performed on an object that "stands for" or represents another object (e.g., using a block for a hamburger). The second category involves use of a doll or other figure as an active agent that can initiate and perform actions independently (putting spoon in doll's hand). Distinguishing between these sequential categories of pre-symbolic and pretend play is important for children with neurodevelopmental disorders at lower levels of cognitive development because they may produce earlier emerging forms of pretend play that could be overlooked with the use of more global play taxonomies.

Over and above the cognitive challenges experienced by young boys with FXS, their phenotypic characteristics (inattention, hyperactivity, repetitive and challenging behaviors) seem particularly likely to interfere with engagement in sustained periods of productive exploratory play with objects and may well impact the emergence of play skills more than for children with other developmental disabilities. In addition to the potential bidirectional effects of limited play skills on cognitive development, it is likely that limitations in object-focused engagement also have a negative transactional impact on opportunities for learning new play skills and for the number of language learning opportunities that young boys with FXS receive from their caregivers.

1.4. Play as a context for the development of language

Although play with objects is important for cognitive development, typically developing children are unlikely to play alone for long periods of time (Kasari et al., 2014). During the period from 9–15-months, play with objects provides a natural context for children to engage in reciprocal interactions with their caregivers, thereby supporting developments in social communication and language (Lifter et al., 2011a).

Through participation in familiar, predictable routines, children learn about the back and forth nature of communication and the ways in which communication acts are paired with object-based exchanges (Halliday, 1984). Thus, children's first communicative turns may take the form of a gesture (e.g., reach or give) produced in response to a cue (e.g., expectant waiting) from the caregiver within a routine with an object. Participation in play-based routines allows children with and without disabilities to coordinate attention between people and objects and increases the frequency of child communication acts directed to the caregiver (Adamson & Chance, 1998; Bakeman & Adamson, 1984).

When children are interested in objects, they are more likely to refer to these objects by producing communication acts for the purpose of initiating joint attention (i.e., labeling/commenting) or behavioral regulation (i.e., requesting), the two most frequently used categories of prelinguistic gestural communication (Wetherby, Cain, Yonclas & Walker, 1988), or by taking a turn with the object (i.e., object exchange; Yoder & Stone, 2006). The rate at which children use these prelinguistic behaviors to communicate intentionally is a significant predictor of later expressive vocabulary in children with mild to moderate developmental delays (McCathren, Yoder, & Warren, 1999). A child who engages with objects infrequently may face a relative disadvantage in the frequency of opportunities to produce gestures for turn-taking, requesting, and referential purposes.

Frequent participation in object-based routines also allows the child to hear consistent linguistic input from caregivers that corresponds to the objects and actions utilized within the routine. When children are productively engaged in play with objects, and even when children produce play actions that are not considered to be symbolic, caregivers can provide verbal language input that follows into the child's focus of attention. Follow-in language allows children to make accurate associations, or mappings, between novel labels and the objects or actions to which these labels refer (Baldwin & Tomasello, 1998). Caregivers of children who spend more time actively and productively engaged in play with objects will have more opportunities to provide a variety of vocabulary words that correspond to the objects in which the child is interested and the actions which the child produces. Conversely, caregivers of children who demonstrate more limited engagement with objects will have fewer opportunities to provide such language facilitating verbal input.

In summary, by supporting time spent jointly engaged, children's object-focused play skills should have a transactional effect on the amount of both follow-in and contingent verbal language input they receive (Removed for blind review, 2010; Removed for blind review, 2006) and in the number of opportunities they have for taking turns and directing communication acts to caregivers. Broadly speaking, joint engagement with objects should facilitate language comprehension (Perryman, Carter, Messenger, Stone, Ivanescu, & Yoder, 2013), the production of nonverbal communication acts (Ninio & Bruner, 1986; Tomasello & Todd, 1983; Williams, 2003), and the transition to verbal communication (Accredolo & Goodwin, 1988).

1.5. Metrics of play with objects

Three metrics have been proposed to represent the association between object play and language development for very young children with developmental delays (Removed for blind review, 2006): (a) frequency of exploratory play actions (i.e., Discriminative Play Actions); (b) play with a variety of different objects (i.e., Object Interest), and (c) production of diverse range of play actions (i.e., Diversity of Play). Although no published studies have examined the relationship between play with objects and later language for children with FXS, a number of studies have considered this association for young children with ASD.

Toth, Munson, Meltzoff, and Dawson (2006), for example, demonstrated that a composite variable representing the frequency of both functional and symbolic play measured at 3–4 years of age was a significant predictor of rate of growth in communication and language as measured by the Vineland between 4 and 6.5 years of age. Similarly, Sigman and McGovern (2005) found that frequency of functional play with objects during early childhood was a significant predictor of long term gains in expressive language for a group of adolescents with ASD.

Adamson, Deckner and Bakeman (2010) demonstrated that lower interest in new objects was significantly correlated with more time spent unengaged for a group of children with ASD at a mean age of 30 months. These authors also found that degree of interest in novel objects was a significant predictor of duration of time spent in supported joint engagement, a triadic engagement state in which parents can provide verbal language input that follows into their child's focus of attention (Adamson, Bakeman, & Deckner, 2004) and which is known to support later language outcomes for children with ASD (Removed for blind review, 2010; Siller & Sigman, 2002). Longitudinally, restricted object interest has been shown to be negatively related the frequency of both responding to and initiating joint attention six months later for a group of toddlers with ASD (Bruckner & Yoder, 2007).

With regard to production of a diverse range of play actions, Yoder (2006) used mixed level modeling to demonstrate that diversity of object play was a significant predictor of growth in lexical density over a one-year time period for minimally verbal toddlers with ASD; that is, the number of different play actions children produced was a significant predictor of rate of growth in the number of nonimitative spoken words children produced one year later. Thus, several metrics, each representing a slightly different aspect of play can be utilized as potential correlates of other child characteristics. Examining patterns of correlations may provide insights into the conceptual underpinnings of play in boys with FXS.

1.6. The present study

The purpose of the current study was to provide a preliminary description of the object play skills of a small group of toddlers with FXS. An additional purpose was to provide an exploratory examination of bivariate associations between standardized and naturalistic measures of cognitive, social communication, and language development and several different metrics of play with objects as measured using the Developmental Play Assessment (Lifter, 2000). We examined four different play variables and specifically hypothesized that frequency of (1) Discriminative Play Actions, (2) Object Interest, and (3) Play Diversity would be positively correlated with chronological age and nonverbal cognitive ability. Additionally, it was expected that these three play variables would be positively correlated with standardized measures of language comprehension and production and negatively correlated with severity of autism symptoms. Finally, the play variables were expected to be positively correlated with measures of nonverbal and verbal child intentional communication collected within a naturalistic context of parent/child interaction. These initial findings have the potential to inform a more complete picture of the behavioral phenotype of young males with FXS. Additionally, improved understanding of the profiles and correlates of object play for this group of children may contribute to the development of more effective interventions targeting developmental domains such as communication and language.

2. Methods

2.1. Participants

Children in the current study were enrolled with their mothers in a larger project testing the efficacy of a parent-implemented communication and language intervention. All child participants were males who met the following inclusion criteria: (a) diagnosis of full mutation fragile X syndrome confirmed through molecular testing; (b) between 2 and 6 years of age; (c) fewer than 10 different spoken words used on a daily basis; and, (d) no uncorrected sensory or motor impairments severe enough to preclude processing and responding to verbal language input. Both items (c) and (d) were according to maternal report. Prior to the implementation of the intervention, a battery of standardized tests, observational assessments, and informant report measures were used to assess each child's communication and language skills and level of cognitive functioning and play skills. All assessments were administered by an experienced masters-level speech/language clinician. Data for the current study were all collected at the pre-treatment assessment. This study was approved by the University's Institutional Review Board.

2.2. Measures

Descriptive characteristics of the participants on the following measures of interest are presented in Tables 1 and 2.

MacArthur-Bates Communicative Development Inventories (MCDI; Fenson, Marchman, Thal, Dale, Bates, & Reznick, 2007). The MCDI is a widely used parent report measure of early vocabulary comprehension and production. All mothers completed the Words and Gestures subscale of the MCDI. Raw scores from the 396-item vocabulary checklist were used as a metric of spoken vocabulary production.

Mullen Scales of Early Learning (MSEL; Mullen, 1995). The MSEL is a standardized measure of cognitive development appropriate for children ages 3 to 68 months. A metric of nonverbal cognitive ability was calculated by combining raw scores from the Fine Motor and Visual Reception subtests of the MSEL, following the approach utilized by Wetherby, Woods, Allen, Cleary, Dickinson, & Lord, 2004), Yoder (2006), and Thiemann-Bourque et al. (2012) Age-equivalent scores are reported to provide a description of the participants.

Preschool Language Scale 5th Edition (PLS-5; Zimmerman, Steiner, & Pond, 2007). The PLS-5 is a standardized developmental language measure appropriate for children from 2 weeks to 6 years of age. Language abilities are assessed in two subscales: Auditory Comprehension and Expressive Communication. Standard and age equivalent scores are reported to provide a description of the participants. Subscale raw scores were used in all analyses.

Autism Diagnostic Observation Schedule (ADOS-2; Rutter, DiLavore, Risi, Gotham, & Bishop, 2012) or *Autism Diagnostic Observation Schedule–Toddler Version* (ADOS-T; Lord, Luyster, Gotham, & Guthrie, 2012) was administered by a trained and research reliable examiner. The ADOS scale and modules were selected based on each child's age and expressive language level as specified in the ADOS manual. To provide a continuous metric of autism symptom severity, ADOS raw scores were converted to calibrated severity scores (Gotham, Pickles, & Lord, 2009). All children in the current study received ADOS module 1 with the exception of one child who was under 30 months of age and to whom a Toddler module was administered. The Toddler module cannot be used for the calculation of a severity score.

Table 1
Descriptive characteristics of participants: standard scores and age equivalents.

| Measures | Participants | | | | | | | | | |
	1	2	3	4	5	6	7	8	9	10
Chronological Age	3.25	2.92	3.33	2.25	3.33	3.58	4.08	3.84	2.58	4.25
Mullen Scales of Early Learning										
Visual Reception Age Equivalent	2.17	2.75	1.42	1.92	2.00	1.92	3.58	2.50	1.42	1.67
Fine Motor Age Equivalent	1.42	1.42	1.17	1.42	1.92	1.83	2.50	2.00	1.33	1.42
Preschool Language Scales 5th Edition										
Auditory Comprehension Standard Score	62	98	50	73	67	78	77	76	76	50
Auditory Comprehension Age Equivalent	1.75	2.67	1.25	1.42	1.92	2.67	3.00	2.58	1.92	1.25
Expressive Communication Standard Score	58	77	50	77	64	64	64	68	75	50
Expressive Communication Age Equivalent	1.17	1.67	0.83	1.42	1.42	1.67	2.00	1.92	1.58	0.75
Total Language Standard Score	57	86	50	74	63	69	69	70	75	50
Total Language Age Equivalent	1.42	2.17	1.0	1.42	1.67	2.17	2.50	2.25	1.75	0.92
Autism Severity Score[a]	4	4	6	–	4	4	7	3	4	10

[a] A severity score of 4 or above indicates symptoms of autism that are frequent and/or severe enough to be considered on the autism spectrum when controlling for CA, IQ, and expressive language (Gotham et al., 2009).

Table 2
Descriptive characteristics of participants: raw scores for standardized assessments.

Participant	Mullen Scales of Early Learning		Preschool Language Scales 5th Edition		MCDI	ADOS Calibrated Severity Score
	Visual Reception	Fine Motor	AC	EC	Words Said	
1	28	19	24	19	1	4
2	33	19	34	25	71	4
3	20	16	19	15	0	6
4	25	19	21	22	33	–[a]
5	26	24	26	22	2	4
6	25	23	34	25	40	4
7	40	29	37	28	65	7
8	31	25	33	27	32	3
9	20	18	26	24	38	4
10	23	19	19	14	0	10

[a] ADOS Toddler Module administered – no severity score available.

2.3. Play assessment procedures

The *Developmental Play Assessment* (Lifter, 2000) entails collecting a video-recorded sample of a child's naturally occurring, unstructured play activities that can be coded and analyzed to determine the frequency of various play actions. The actions are then organized to determine the frequency and variety of actions within a sequential set of developmentally organized play categories. In the adaptation of the DPA used in the current study, three sets of toys were individually presented to the child by an examiner. Each toy set was placed on the floor in the proximity of the child and the child was allowed to explore that set for 4 min. The examiner showed the toys to the child ("Look, you can play with my toys!") but did not model any play actions. The parent was not in the room during the DPA session. Session videotapes were coded to derive the play variables of interest.

Parent/Child Play Sessions were conducted to serve as baseline sessions for the language intervention project. For each 10-min baseline session, the mother was given three developmentally appropriate toys and was instructed to play with her child as she usually would. A new toy set was provided for each baseline session. Partial interval coding was used to code instances of nonverbal and verbal communication acts from each videotaped session. A composite variable was then derived by computing the average frequency of both types of child communication acts across all baseline sessions for each participant.

2.4. Coding of play variables

The procedure for coding of play behaviors was adapted from the instructions for scoring the Developmental Play Assessment as described by Lifter (2000). Although the use of 15 play categories was originally specified, the current study utilized 11 play categories distributed into 8 play levels following Thiemann-Bourque et al. (2012) (Table 3). Previous research has revealed that actions included in the four excluded play categories, which involve object substitution and other higher order symbolic skills, are infrequently produced by children with developmental disabilities (Lifter, Mason, & Barton, 2011).

DPA administrations were event coded using Playcoder (Tapp & Yoder, 2003), a software program that automates the coding of play behaviors from video-recordings of the DPA. Playcoder enables a coder to select a toy from the list of toys used during the play assessment and then select from a list of anticipated actions that could be performed with that toy. The list of anticipated actions includes both undifferentiated and differentiated play acts. Undifferentiated actions (e.g., mouthing, shaking, and banging) do not provide evidence that the child understands what to do with a toy. Differentiated actions, in contrast, are play action that demonstrate some understanding of the properties of a given toy (Lifter, 2000, Lifter, Sulzer-Azaroff, Anderson, & Cowdery, 1993). Play sessions were coded by two graduate students who were trained by the first author. Raw scores for the DPA play categories are also displayed in Table 3.

The four summary metrics of play utilized in the current study were frequency counts of: Indiscriminate Play Actions, Discriminative Play Actions, Object Interest, and Play Diversity. Indiscriminate Play Actions were defined as mouthing, or repetitive sensorimotor manipulation (including shaking and banging). Discriminative Play Actions was defined as the total number of non-imitative, differentiated play actions (i.e., purposeful manipulation of a toy) produced during the DPA session. Object Interest was defined as the number of toys on which children used differentiated play actions. Play diversity was defined as the number of unique differentiated play actions that were produced during the DPA session. Scores for these summary level play variables are displayed in Table 4.

2.4.1. Coding of child communication acts

A 5-s partial interval coding system was used to identify child acts of nonverbal and verbal intentional communication based upon definitions adapted from the Communication and Symbolic Behavior Scales: Developmental Profile (Wetherby &

Table 3
Raw Scores for Developmental Play Assessment Categories.

Play Level	Play Category (and Definition)	Participant									
		1	2	3	4	5	6	7	8	9	10
1	*Indiscriminate Play Actions*										
	Mouthing, Shaking, Banging (All objects treated alike)	31	18	51	41	42	56	86	41	120	55
2	*Discriminative Play Actions*										
	Actions on single objects (Differentiates among objects)	4(3)	31(4)[a]	5(2)[b]	11(1)	75(3)[b]	5(3)[b]	6(3)[b]	5(2)[b]	5(3)[b]	16(2)[b]
	Take Apart Combinations (Separates configurations of objects)	17(8)[a]	18(6)[a]	16(3)[b]	7(5)[b]	16(6)[a]	21(9)[a]	18(6)[a]	43(8)[a]	24(8)[a]	12(5)[a]
3	*Combinatorial Play Actions*										
	Presentation Combinations (Recreates configurations of objects)	11(5)[a]	44(4)[a]	24(4)[a]	5(4)[b]	9(2)[b]	6(3)[b]	12(3)[b]	31(4)[a]	10(3)[b]	10(4)[a]
	General Combinations (Creates simple nonspecific configurations)	1(1)	9(2)[b]	5(1)	1(1)	8(3)[b]	6(4)[2]	0	3(2)	0	11(3)[b]
	Pretend Self (Relates conventional object to self with familiar action)	4(2)	2(2)	0	0	0	1(1)	1(1)	0	0	0
	Specific Physical Combinations (Combines objects based upon unique physical characteristics)	6(2)[b]	0	1(1)	0	6(2)[b]	3(1)	2(1)	0	1(1)	1(1)
4	*Pretend Play Actions*										
	Child-as-Agent (Extends familiar actions to dolls or figures)	8(2)[b]	3(2)	0	0	0	1(1)	0	4(2)	2(2)	0
	Specific Conventional Combinations (Combines objects based on conventional uses)	0	0	0	0	0	0	0	0	0	0

[a] Mastered – 10 occurrences with at least 4 action types.
[b] Emerging – 5 occurrences with at least 2 action types.

Prizant, 2002). Verbal communication acts included both spontaneous and prompted words and phrases produced by the child. Nonverbal communication acts included gestures (reaches, gives, shows, points, conventional signs) as well as vocalizations accompanied by eye contact. ProcoderDV (Tapp, 2003), a software program enabling observational coding from digital video-recordings, was used to code each 10-min baseline session. Data were extracted from baseline sessions administered to all participants at the pre-treatment. Coded files were exported into MOOSES software (Tapp, Wehby, & Ellis, 1995) for calculation of the cumulative frequency of intervals during which a communication act occurred. Frequencies were averaged across baseline sessions to obtain a composite for each variable. Four participants (1, 4, 7, and 9) had 5 baseline sessions, three participants (2, 5, and 10) had 8 baseline sessions, and three participants (3, 6, and 11) had 11 baseline sessions.

2.5. Reliability

For the DPA, inter-rater agreement was computed by having a second coder independently recode 20% of the DPA sessions, which were randomly selected. The primary coder trained the reliability coder through coding of practice videos and a series of consensus discussions. Interobserver reliability was computed using intraclass correlation coefficients which reflect the proportion of the variability in the reliability sample that is due to between-participant variance relative to the proportion of variance that is due to differences between coders (Shavelson & Webb, 1991). Intraclass correlation coefficient values of 0.6 are considered acceptable. Reliability between the two coders was .77, .89, .85, and .90 for Indiscriminate Actions, Discriminative Actions, Object Interest, and Play Diversity, respectively. For the language variables, this same process was used and yielded intraclass correlation coefficients of .87 and .93 for nonverbal and verbal communication acts, respectively.

2.6. Analysis plan

Kendall's *Tau B* was utilized in all analyses as the index of association given its suitability for small sample sizes. Test values for this statistic range from 1 to −1 and reflect the degree of concordance or discordance between ranked data; that is, the difference between the probability that the observed data for two variables are in the same order *versus* the probability that the data for these same variables are not in the same order. One tailed *p*-values were used, as directional hypotheses

Table 4
Raw scores for coded play variables.

Play Variables	Participants									
	1	2	3	4	5	6	7	8	9	10
Object Interest	11	7	5	5	6	13	9	8	10	5
Diversity of Play Actions	23	20	11	11	16	22	14	18	17	15
Discriminative Play Actions	51	107	51	24	114	43	39	86	42	50
Indiscriminate Play Actions	31	18	51	41	42	56	86	41	120	55

were made for all analyses. Given the developmental level of the participants, most would be expected to score at floor levels when using standard scores in an analysis. Thus, raw scores were used to represent performance on the MSEL and PLS-5.

3. Results

3.1. Descriptive analysis of object play skills

The first analysis examined the distribution of play levels that were either Mastered or Emerging according to Lifter's (2000) criteria; that is, a play level is considered Mastered if a child produces a minimum of 10 actions of at least 4 different types. A play level is considered Emerging if a child produces a minimum of 5 actions of at least 2 different types. Only one child of the ten participants achieved mastery of Discriminative Play Actions, whereas this category was emerging for 7 participants. In contrast, 8 children had mastered Take Apart Combinations, with this category emerging for the remaining 2 participants. Five children demonstrated mastery of Presentation Combinations, with this category emerging for the remaining 5 participants. This was the highest level of play that was either emerging or mastered for all 10 participants. General Combinations and Specific Physical Combinations were emerging for 4 and 2 participants, respectively. The Pretend Self category was not emerging for any of the participants and, interestingly, only 8 actions (4 different) were produced in this category across all 10 participants. Finally, Pretend Play Actions were observed for only one participant who produced two different actions in this category.

3.2. Associations between play and other child characteristics

None of the play variables was significantly associated with chronological age or nonverbal cognitive developmental level. Object interest (i.e., the number of different toys touched) during the DPA session was significantly and positively associated with both the Auditory Comprehension and Expressive Communication subscales of the PLS-5, $Tau\ b = .476$, $p < 033$, and $Tau\ b = 424$, $p < .050$, respectively, both large effect sizes. The positive association between Object Interest and parent report of spoken vocabulary, $Tau\ b = .326$, $p < 101$, and the negative association between Object Interest and autism severity, $Tau\ b = -.365$, $p < .101$, failed to reach significance. There was a significant and negative association between Diversity of Object Play (i.e., number of different action types) and severity of autism symptoms, $Tau\ b = -.497$, $p < .042$, a large effect size. There were no significant associations for either Discriminative or Indiscriminate Play Actions.

3.3. Associations between play and child communication acts

The next analysis examined associations between the play variables and two composite measures of child intentional communication. Object Interest was significantly and positively correlated with the number of nonverbal (i.e., prelinguistic) communication acts that children directed to their mothers, $Tau\ b = 698$, $p < .003$, a large effect size. Similarly, there was a significant and positive association between Play Diversity and number of nonverbal communication acts, $Tau\ b = .432$, $p < .043$, a similarly large effect size. There were no significant associations between Indiscriminative or Discriminative Play Actions and either of the variables representing child intentional communication.

4. Discussion

The purpose of this study was to provide an initial description of the object play skills of male toddlers with FXS. We first examined the play levels that were considered to be Mastered or Emerging according to the Developmental Play Assessment (Lifter, 2000). Play Level 2, Discriminative Play Actions, includes both Actions on Single Objects and Take-Apart Combinations, both of which should emerge at a developmental level of 7–9 months. In this level, 7 of 10 children had mastered Take-Apart Combinations (separating objects from the configuration in which they are presented). In contrast, only 1 child had mastered Actions on Single Objects (performing a single action which preserves the physical or conventional characteristics of the object), whereas this category was emerging for 7 other children. Take-apart actions can be considered more structured than single discriminative actions, in that there are not a variety of different play options available when separating an object that is presented within a set configuration. For example, the action of taking a piece out of a puzzle or taking a cup out of a stack of nesting cups is constrained by the physical arrangement of the objects. In contrast, when picking up a single object, such as a ball, the child must make an intentional decision about what to do with it (roll it on the floor, kick it, put it in a truck, try to bounce it, etc.). It was our clinical observation that the participants in the study often used the available objects in stereotyped ways; that is, spinning plates, spinning pegs, running trains back and forth on a table), which would also account for the relative delays in mastering the category of Single Actions on Objects.

Within DPA Play Level 3, Combinatorial Play Actions, Presentation Combinations should emerge at a developmental level of approximately 9 months. This play level was mastered by 5 children and emerging for the other 5 children. As with Take Apart Combinations, Presentation Combinations includes a variety of fairly structured actions as the objects are reassembled into combinations constrained to some extent by their physical characteristics (e.g., putting pieces into a puzzle, stacking rings on a post, stringing beads). In fact, we often observed children to repetitively perform take-apart actions alternated with actions that reassembled the initial configurations of the objects (e.g., taking pegs out of pegboard and putting pegs

back in; taking pieces out of puzzle and putting them back in). Even though some of these play actions have a repetitive quality, children did get credit for taking-apart and combining the objects into and out of their original configurations. These action patterns are not necessarily productive relative to children learning because, as suggested by Roeyers and van Berckelaer-Onnes (1994), spending prolonged amounts of time engaged in stereotyped, self-stimulatory, and inflexible behavior patterns may restrict experience with objects, limit the capacity to explore objects, and learn what other actions objects can perform, or what is conventionally done with objects. It seems likely that this is what the child participants in our study were experiencing.

Presentation Combinations, in Level 3, was the highest play category for which all children demonstrated either mastery or emergence of play actions. General combinations, also in Level 3, should emerge at a developmental level of 12 months and were emerging for 4 of the 10 participants in the current study sample. This play category includes actions that involve at least two objects and create simple nonspecific combinations, including the spatial relationships of containment and support (putting pegs into a bowl, lining up animal figures) (Bowerman, 1996).

Play Level 3 also includes Pretend-Self actions, which should begin to emerge between 13–20-months, developmentally. These actions, in which the participant directs conventional actions with an object to himself or performs a conventional gesture with an object, were observed for only 4 of the 10 participants (e.g., bring spoon to mouth, bring cup to mouth, comb hair, put bowl on head as hat). Of these participants, only 2 participants produced more than one action type. Thus, Pretend-Self actions were not emerging or mastered for any of the participants. Lack of such functional play actions may be attributed to limited engagement in play and caregiving routines for the current participants, which would lead to fewer opportunities to see caregivers demonstrate the conventional uses of objects. In fact, most of the mothers reported to us that they hurried through caregiving routines as quickly as they could in light of their child's challenging behaviors and noncompliance. Similarly, few of these caregivers reported being able to sit down and engage their child in back-and-forth routines with objects. In our clinical opinion, this pattern of caregiving was due to a variety of behaviors including inattention and escape- and tangibly-maintained challenging behaviors.

Finally, in Level 3, only 2 participants produced any Specific Physical Combinations. These play actions, which should emerge at approximately 15 months (Belsky & Most, 1981), included stringing beads and putting pegs in a pegboard for both participants. Only 1 participant produced any play actions that could be considered to be symbolic (i.e., Child-as-Agent) and did this by directing two different actions to the doll (combing the doll's hair – 3 times; putting the spoon to the doll's mouth – 5 times). These types of play actions should emerge at a cognitive level sometime after 20 months.

Children in this study had developmental levels between 17 and 43 months according to the Visual Reception subtest of the Mullen Scales of Early Learning. Their fine motor skills were slightly behind their nonverbal developmental levels and ranged from 14 to 30 months. It is possible that fine motor skills were underestimated if children did not want to engage with manipulatives or were not able to attend long enough to comply with task demands. However, based upon DPA performance, it appears that participants' play skills were delayed relative to both their nonverbal developmental level as well as their fine motor ability. With regard to nonverbal cognitive skills, 9 of 10 participants achieved a developmental level at which Child as Agent play actions should have been emerging. Similarly, with regard to fine motor skills, 5 of 10 participants achieved a developmental level at which Child as Agent play actions should have been emerging. Yet, only one child produced any Child as Agent play skills and this child produced only two different actions. It should be mentioned, however, that all children seemed to be progressing in play according to a typical developmental sequence. That is, play skills were delayed in emergence but did not emerge in an atypical order.

We also examined the bivariate associations between the metrics of play with objects and the developmental characteristics of the participants. Counter to our expectations, none of the play variables were significantly associated with chronological age or nonverbal cognitive developmental level as represented by a composite variable summing across Visual Reception and Fine Motor raw scores on the Mullen. Object Interest was a significant correlate of both language comprehension and production as measured by a standardized assessment, the PLS. Although not reaching significance, the positive association between Object Interest and parent report of spoken vocabulary and the negative association between Object Interest and severity of autism symptoms were in the hypothesized direction. However, a study with a larger sample size would be needed to support or refute these potential findings. As predicted, there was a significant and negative association between Diversity of Object Play (i.e., number of different action types) and severity of autism symptoms. In contrast to our expectations, however, we failed to find any significant associations between Indiscriminate or Discriminative Actions on Single Objects and child characteristics.

This differential pattern of findings raises the question of why some of the play metrics were relatively more effective in demonstrating an association with child characteristics. It would appear that metrics that captured interest in a variety of objects or diversity of play actions seemed to be more effective in demonstrating an association with standardized measures of child communication and language. According to Kasari and Chang (2014), frequency of play is less important than the diversity of play actions produced because children may have a high frequency of producing the same action over and over; that is, a higher measured frequency of play actions may be due to numerous repetitive play acts whereas play diversity indicates the ability to engage in more flexible and varied play. This hypothesis gains some support from our observations that children often did not produce very many individual actions on objects when these actions were not constrained by a pre-existing structure. Additionally, we often observed children to be repeating actions such as spinning during the DPA sessions. Object Interest may capture an important facet of play as a child who is open to examining and playing with a variety of objects may be easier to engage in a play routine or participation in a sustained period of triadic joint attention.

If early exploratory play has an information gathering function (Williams, 2003), then children who are not engaged and interested in a variety of objects or who use objects in a repetitive fashion, will miss out on recurring opportunities to learn about objects. Similarly, the negative correlation we observed between Play Diversity and autism symptom severity may reflect either a lack of interest in toys or the use of toys in a repetitive manner.

Finally, we examined associations between the three metrics of play and the frequency of nonverbal and verbal acts of intentional communication that were produced within play-based interactions with each child's mother. Our analyses revealed significant concurrent associations between the frequency of nonverbal communication acts with both Object Interest and Play Diversity, confirming the findings of Yoder and Lieberman's recent meta-analysis (Lieberman & Yoder, 2012). In addition, Bruckner and Yoder (2007) found that, when controlling for initial play level, restricted object use in young children with ASD was inversely related to several important social communication skills, such as response to joint attention and the ability to coordinate attention between object and person. To coordinate attention a child must be interested in both people and objects. To the extent that a child is not interested in objects, they may not be motivated remain engaged in a play routine or to share affect with a communicative partner. Similarly, a child who is not interested in objects may be unlikely to direct communication acts (i.e., comments or requests) to a play partner or to take an object-exchange turn within a play routine. Our failure to detect an association between any of the play variables and the frequency of verbal communication acts may be attributed to the limited instances of verbal communication acts that were produced by child participants in the current study.

4.1. Limitations

The current study had several limitations including the small sample size and the concurrent correlational design. A larger sample would have provided more power to detect significant associations. Additionally, use of a longitudinal design and controlling for initial levels of nonverbal and verbal communication would have lent support to the conclusion that the play variables are causally related to later receptive and expressive language levels. Our clinical impression was that the participants in this study were challenged by high levels of social anxiety, although this was not formally measured. Most of them had difficulty not only being separated from their mothers but also having an unfamiliar examiner in the room. This may have negatively influenced the results of cognitive and language testing. Furthermore, the Developmental Play Assessment was administered by an examiner; it is possible that more frequent or diverse play actions or higher developmental levels of play might have been observed if a caregiver/child format had been used.

4.2. Clinical implications and future directions

Productive play with objects provides children with a point of entry into naturalistic exchanges with communicative partners. These interactions support the subsequent development of more advanced play skills as well as other pivotal skills related to social communication and language (i.e., turn-taking, prelinguistic intentional communication, verbal language comprehension and production). Results of the current study suggest that increased participation in play-based routines with objects is an important intervention goal for young children with FXS, as these abilities seem to be especially delayed relative to levels of nonverbal cognitive development. Supporting the development of such routines is an especially relevant goal given the presence of phenotypic characteristics that are likely to negatively impact the ability to sustain periods of productive play with objects as well as the ability to interact with social partners. These behaviors include hyperarousal, inattention, repetitive object use, and escape- and tangibly-maintained challenging behaviors. Thus, interventions for young children with FXS should focus on establishing the regular use of back-and-forth play and caregiving routines with objects in which parents can provide modeling of more advanced play, opportunities for turn-taking, communication prompts, follow-in language, and contingent responses to child communication acts. For example, after learning the routine of rolling a ball back and forth, the child may initiate the routine (e.g., by bringing a ball to the caregiver) or take a turn in an ongoing routine (e.g., rolling the ball back to the caregiver). In addition, the caregiver can use strategies, such as expectant waiting, to provide the child with opportunities to request that the routine be continued. Following a child's prelinguistic comment or request, the caregiver can use a noun, verb, or function word to put the presumed meaning of the child's communication act into words (i.e., linguistic mapping of the child's reaching gesture: 'You want the ball!'). Many types of objects can be incorporated into a routine that includes a playful, turn-taking interaction in which the child and caregiver share a common focus of attention. Importantly, these types of interactional routines can also be established for daily caregiving activities, such as bath time and getting dressed (Wilcox & Woods, 2011), providing an additional naturalistic context for the development of communication and language.

In conclusion, future studies of the development of play in fragile X syndrome should include a larger sample, a typically developing comparison group, as well as children with other forms of intellectual disability to provide insights into whether observed patterns of play are related to limitations in cognitive level or to other aspects of the behavioral phenotype of FXS. Additionally, the inclusion of a comparison group of girls with FXS would also reveal any gender specific aspect of play performance. Finally, there is ongoing speculation as to whether autism symptoms in FXS represent the same underlying psychological and neurobiological impairments as in nonsyndromic ASD (Removed for blind review, in press). A comparison group of boys diagnosed with ASD would provide information as to whether developmental accomplishments in play are influenced by autism symptoms in the same way across both disorders.

Appendix A. Continuing education

CEU Questions

1. The behavioral phenotype of young males with FXS includes:
   a. Cognitive delays
   b. Inattention and hyperactivity
   c. Repetitive behaviors
   d. Symptoms of ASD
   e. All of the above
2. Putting the pieces back into a puzzle can be considered:
   a. A functional play action
   b. A general combination
   c. A specific combination
   d. A discriminative play action
3. In this study, the number of nonverbal communication acts children produced during a naturalistic interaction with their mother was significantly correlated with which of the following DPA variables:
   a. The number of toys children touched
   b. The number of actions children produced
   c. The number of different action types children produced
   d. The number of indiscriminate actions children produced
   e. A and B
   f. A and C
   g. A and D
4. A higher interest in new objects should result in more time spent:
   a. In producing discriminative play action
   b. In producing Take-Apart combination
   c. In Supportive Joint Engagement
   d. Responding to parent's attentional bids
5. Children's early participation in interactional routines with objects is related to their:
   a. Ability to produce pretend play actions
   b. Ability to produce discriminative play actions
   c. Ability to produce spoken words
   d. Ability to produce nonverbal communication acts

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