Sensory Processing Correlates of Occupational Performance in Children With Fragile X Syndrome: Preliminary Findings

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KEY WORDS
- developmental disabilities
- sensory integration
- sensory modulation

OBJECTIVES. This preliminary study examined sensory processing and its relationship to occupational performance in children with fragile X syndrome (FXS) to guide research and evidence-based practice.

METHOD. Fifteen school-aged boys with full-mutation FXS were assessed with three occupational performance measures (School Function Assessment, Vineland Adaptive Behavior Scales, play duration) and three sensory processing measures (Sensory Profile, Tactile Defensiveness and Discrimination Test–Revised, Sensory Approach–Avoidance Rating). Data were analyzed using partial correlation procedures.

RESULTS. Several significant correlations were found, independent of effects of age and IQ. Avoidance of sensory experiences (internally controlled) was associated with *lower* levels of school participation, self-care, and play. Aversion to touch from externally controlled sources was associated with a trend toward *greater* independence in self-care—opposite of expectations.

CONCLUSION. This study links sensory processing vulnerabilities with individual differences in occupational performance and supports a dynamic view of self-organizing systems. Children's uses of avoidant versus independent behaviors may reflect different self-regulatory or coping strategies that potentially mediate the relationship between sensory processing deficits and occupational behaviors and warrant further investigation.

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Fragile X syndrome (FXS) is an X-linked disorder that is the most common inherited cause of mental retardation. It affects persons of all races and ethnic groups, with an estimated frequency at 1 in 4,000 males and 1 in 8,000 females (Crawford et al., 1999; Turner, Webb, Wake, & Robinson, 1996). The gene FMR-1 (Fragile X Mental Retardation-1) is located on the X chromosome, and a mutation in this gene causes FXS. The genetic code for this gene normally contains between 5 and 50 repetitions of CGG (cytosine-guanine-guanine) sequences, the average being around 30 (Bailey, 1997). Some persons may have an expansion of 50 to 200 CGG repeats and are referred to as premutation carriers. Although many persons with the premutation may show no effects of the disorder (Mazzocco & Holden, 1996), mounting evidence shows that others are affected (Hagerman, 1996). Persons with full-mutation FXS are those having 200 or more CGG repeats. In most cases, an expansion of this size results in decreased production of the FMR protein assumed to be essential for normal brain functioning.

A variety of unusual behaviors (e.g., tactile defensiveness, gaze aversion, hyperactivity, hyperarousal, hand flapping) thought to reflect difficulties in sensory processing are observed clinically in children with FXS (Scharfenaker et al., 1996; Shopmeyer & Lowe, 1992; Stackhouse, 1994). Occupational therapists often receive referrals for children with FXS to mitigate sensory processing difficulties and help families cope more effectively. Anecdotal reports based on expert clinical

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observations add to our understanding of the potential impact of sensory processing difficulties on occupational performance; however, empirical evidence is lacking.

Within the field of occupational therapy, one study (Baranek, Hooper, Hatton, & Bailey, 2002) documented the nature of sensory processing disruptions in boys with FXS and its association with behavioral problems. Miller et al. (1999) and Miller and McIntosh (1998) found that children with FXS manifested the most severe sensory processing disorders of all the clinical groups they studied (e.g., autism, attention deficit hyperactivity disorder). Researchers from fields outside of occupational therapy have provided the bulk of the scientific literature about FXS (e.g., Bailey, Hatton, & Skinner, 1998; Belser & Sudhalter, 1995; Boccia & Roberts, 2000; Cohen, 1995; Hatton, Bailey, Hargett-Beck, Skinner, & Clark, 1999; Warren & Nelson, 1994). These important studies document physical and behavioral features, genotypic and phenotypic correlates, developmental profiles, or consequences of physiological hyperarousal. Unusual sensory responses are described occasionally as aspects of temperament (Bailey, Hatton, Mesibov, Ament, & Skinner, 2000) or a consequence of physiological hyperarousal (Cohen, 1995) but are not targeted for study specifically.

Occupational performance of persons with FXS has not been investigated directly. A few studies that address the consequences of FXS on adaptive behavior (Dykens et al., 1996; Freund, Peebles, Aylward, & Reiss, 1995) focus on developmental changes, noting that age correlates significantly with increases in adaptive skills during the preschool years and that adaptive skills often surpass cognitive performance on standardized tests as boys with FXS approach adulthood. None of these studies specifically relates sensory processing to individual differences in occupational performance.

Few conceptual models guide interventions in this area. Assumptions of linear causality are evident in some practices (i.e., remediation of sensory processing deficits), whereby impairments in one subsystem (sensory processes) are thought to directly affect occupational performance and guide the therapist's treatment accordingly. However, newer theories using dynamical systems perspectives (Thelen, 1989; Thelen & Smith, 1994) highlight interactions among various intrinsic capacities and stress the importance of self-organizing functions that partially depend on performance in contextually relevant tasks. Thus, a less direct relationship between components (i.e., sensory processing) and occupational performance is implied in the newer perspective. This perspective offers insights into alternate interventions, such as providing task and environmental modifications to support engagement in occupation.

The purpose of this preliminary study was to investigate whether sensory processing functions are associated with occupational performance in children with FXS. Based on our review of the literature, we hypothesized that children with higher levels of sensory processing vulnerabilities would present with greater challenges in occupational performance across areas of school function, self-care, and play. Findings generated from this study will be used to guide further research and evidence-based practice.

Method

This study used a correlational design to measure the association between sensory processing and occupational performance. Descriptive methods also were used.

Participants

Fifteen boys with full-mutation FXS, ranging in age from 53 to 126 months (M = 93 months, SD = 22), participated in this study. The boys were recruited through a large, multistate research project studying the developmental trajectories of children with FXS. Genetic testing (blood samples) confirmed full mutation for each child. Only boys were recruited for this study because boys are typically more severely affected than girls. Informed consent was obtained from the all the children's parents. The children received a toy; their parents received financial remuneration, including travel expenses.

Race or socioeconomic status was not restricted for this study. Thirteen (87%) children were Caucasian, and 2 (13%) were African American. Two families received public assistance. Educational levels of the primary caregivers (mothers) were reported. Three (20%) mothers were high school graduates; 7 (47%) attended some college courses; and 5 (33%) were college graduates. Data on the fathers were not collected because in some cases fathers were not caregivers or providers.

The children had a mean Brief IQ Composite Score in the mild range of mental retardation (M = 60, SD = 14.29) on the Leiter International Performance Scale–Revised (Roid & Miller, 1997), a nonverbal measure of cognitive skills. Considerable variability existed in cognitive performance (score range = 42–93). Seven children were functioning 3 to 4 standard deviations below the normative mean of the Leiter standardization sample. Three children's scores fell 2 to 3 standard deviations below the normative mean. Only 1 child was functioning within the average range on this standardized test. (One child was untestable.) As a group, the four subtests from the Visualization and Reasoning Battery comprising the "Brief IQ" reflected little variation, with child scores aver-

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aging about 2 standard deviations below the normative mean for all four subtests.

Measures

Assessment of sensory processing components. Few well-standardized tools exist to measure sensory processing functions in a population of children with mental retardation. Furthermore, different formats (parent reports, standardized behavioral tests, naturalistic observations) yield contrasting information about sensory processing abilities (Baranek, Foster, & Berkson, 1997). Thus, we opted to use a comprehensive, multimodal assessment process. To tap these functions, we used one parent-report measure (Sensory Profile [Dunn, 1999]) and two observational measures (Tactile Defensiveness and Discrimination Test–Revised [TDDT-R; Baranek, 1997] and Sensory Approach–Avoidance Rating).

The Sensory Profile is a 125-item parent questionnaire that inquires about a child's responses to sensory events in his or her daily environment (e.g., Is your child bothered by loud noises?). It has been validated for use with school-aged children but does not provide norms for children with FXS, specifically. The items are scored on a 5-point Likert scale ranging from always (1) to never (5). Lower scores on this assessment indicate greater problems in processing sensory information. We used two scores in our analyses: (a) total score for the Short Sensory Profile (SSP; 38 items) and (b) total score of Group 1 items (auditory, visual, vestibular, touch, multisensory, and oral sensory processing categories [total of 65 items]) from the original research version of the Sensory Profile. The SSP provides published cut-off scores. Scores at or above 155 points are considered "typical"; scores from 154 through 142 are reported as a "probable difference" (1 standard deviation below the mean for the typical reference sample); and scores at or below 141 are reported as a "definite difference" (2 standard deviations below the mean for the typical reference sample). Reliability (Cronbach's alpha) of the SSP total score ranges from .90 to .95 for published reference samples (Dunn, 1999).

The TDDT-R is a standardized behavioral assessment measuring tactile processing that has been validated previously for use with children with developmental disabilities between the ages of 3 and 12 years (Baranek & Berkson, 1994; Baranek et al., 1997). The tool has been used reliably in these studies to discriminate levels of sensitivity (aversive responses) to tactile stimuli, an indication of construct validity. Total scores are derived for each of two scales: *Externally controlled* tactile experiences (responses to stimuli such as stickers or a finger puppet applied by the examiner) and *internally controlled* tactile experiences (responses during child-initiated exploration of tactile media, such as lotion, dried noodles, or sand). Higher scores on the TDDT-R reflect greater difficulties with tolerating tactile experiences. Interrater reliability was reassessed and is reported later within this section.

The Sensory Approach–Avoidance Rating was developed for this study as an observational measure of sensory defensiveness in a more naturalistic context of interaction with novel toys. Item selection was based on current conceptual models of sensory processing (Dunn, 1997; Baranek, Reinhartsen, & Wannamaker, 2000) and evidence from a review of the empirical and clinical literature indicating that children with sensory defensiveness are less tolerant of sensory experiences across modalities and less likely to engage with materials that encompass such properties (Ayres, 1979; Dunn, 1997; Kimball, 1993; Kinnealey, Oliver, & Wilbarger, 1995). Toys were chosen for their novelty as well as for their inherent *multisensory* properties with which the child could interact spontaneously. These methods provided preliminary construct and face validity for use of such a measure in this study.

For the Sensory Approach–Avoidance Rating, the children were presented with nine novel multisensory toys to explore individually. The specific toys were a water log, porcupine fish, neon-colored molding sand, rain stick, switch-activated fan, car with sound–light activation, kaleidoscope, spinning board, and large blue ball. Although the research team classified the toys on the basis of their sensory properties for descriptive purposes, it should be emphasized that we were interested in the *multisensory* nature of the experiences. In fact, all children's toys available commercially inherently exhibit some level of tactile and visual properties, and it is impossible to isolate sensory features of toys in naturalistic observations.

All toys contained a minimum of three interactive sensory features. For example, the water log provided a smooth and slippery tactile experience and contained bright-blue water and glitter that reflected the light and attracted visual inspection. When squeezed, the toy elicited an audible sound. The classifications for the sensory properties were derived by research team consensus (100% agreement across independent raters for the most salient sensory properties of the toy). The primary features of the toys were three tactile, three auditory, one visual, and two vestibular. The secondary sensory features were six visual, two tactile, and one auditory. The tertiary sensory features were four auditory, three tactile, and two visual. A variety of sensory properties were represented in this task, and all toys contained a minimum of three sensory features. Gustatory–olfactory features were not included in this study because they rarely occur in toys designed for school-aged children. The child's level of approach or avoidance for

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each toy was observed and rated on a 3-point scale (0 = engages–approaches, 1 = approach–avoidance behavior, 2 = avoids engagement–aversion) for each toy. The sum total across all novel sensory toys was used for analysis. Interrater reliability is reported later within this section. This measure differs from the TDDT-R in that it measures level of engagement during free-play exploration of novel *multi*sensory toys rather than level of discrete responses to structured tactile stimuli. Like the TDDT-R, higher scores indicate greater problems tolerating sensory experiences.

Assessment of occupational performance. Three domains were selected to assess occupational performance: self-care, school function, and play. Measures included the Vineland Adaptive Behavior Scales–Daily Living Skills (VABS; Sparrow, Balla, & Cicchetti, 1984), the School Function Assessment (SFA; Coster, Deeney, Haltiwanger, & Haley, 1998), and a measure of play duration with the toys.

The VABS is an interview-based survey administered to the parents of children birth through 18 years of age. It assesses the child's proficiency and independence in performing various daily activities across several domains (social, daily living, communication, motor). Only the Daily Living Skills section was used in this study because the occupational performance domain of interest was self-care and not overall adaptive functioning. Standard scores were used in the analyses. Reliability of the VABS is reported to be .93 to .99 (Sparrow et al., 1984).

The SFA is a questionnaire that obtains reports of a child's functional performance on school-related activities compared with his or her same-age peers. The classroom teacher was used as the informant for the children in this study. We derived three quantitative measures of "school functioning": (a) the sum score of Part I: Participation (the level of the child's participation summed across 6 school settings: classroom, playground, transportation, bathroom, transition, meals); (b) the sum score of all criterion scores in Part III: Activity Performance–Physical Tasks (12 physical tasks, such as maintaining and changing positions, using materials, eating and drinking, and going up and down stairs); and (c) the sum score of all criterion scores in Part III: Activity Performance–Cognitive/Behavioral Tasks (9 tasks such as functional communication, following social conventions, task behavior and completion, and behavioral regulation). Internal consistency reliability is reported as .92 to .98; test–retest coefficients ranged from .82 to .98 for the reference samples (Coster et al., 1998).

The third occupational performance category—play duration—was a measure of the amount of time the child *engaged* (i.e., physically interacted) with the novel sensory toys provided during the semi-naturalistic play assessment in our laboratory. The total time playing was calculated in whole seconds (to a maximum of 5 min) for each toy and then summed across all multisensory toys for use in the analyses. This measure is not meant as a comprehensive or generalizable measure of play but, rather, as a snapshot of one play context with novel toys. Amount of time attending to or interacting with materials has been documented as an important component of engagement and participation (McWilliam, Trivette, & Dunst, 1985); such measures have been used previously with children with developmental disabilities (Case-Smith & Bryan, 1999; McWilliam & Bailey, 1995; Stone, Lemanek, Fishel, Fernandez, & Altemeier, 1990). The importance of physical exploration of toys as a basis for development of higher level play skills (Belsky & Most, 1981) and participation in joint interactions with caregivers (Bakeman & Adamson, 1984) also is well documented.

Interrater Reliability

Interrater reliability estimates for the TDDT-R, Sensory Approach–Avoidance Rating, and play duration were established by two independent raters from videotaped observations of 3 children (20% of sample). On the TDDT-R, above 90% agreement was obtained. Agreement was 98% for the Sensory Approach–Avoidance Rating and play duration measures.

Procedure

These measures were administered as part of a larger research protocol that included a full 2-hr occupational therapy evaluation and a cognitive assessment. The occupational therapy and psychological evaluations were conducted in a counterbalanced order to minimize fatigue effects across measures. All sessions were videotaped for later coding on the following measures: TDDT-R, Sensory Approach–Avoidance Rating, and play duration. Assessment was done either on site (a small carpeted clinic room at the university) or in a small room in the child's school or home. Unnecessary distractions were minimized across environments. Families brought the child's favorite toys to ease the transition and increase comfort in a new environment. Parents or a familiar adult were present during the initial free-play until the child was comfortable with the examiner. When the child's comfort level seemed appropriate, the caregiver left the room, and the child remained with one examiner and one videographer. Breaks and access to caregivers were provided as needed.

One parent (in most cases, mother) for each child filled out the Sensory Profile and was interviewed about self-help skills on the VABS by a trained examiner (psychologist or educational specialist). The SFA was completed by the child's classroom teacher. Because teachers usually were not present on the day of the child assessments, the SFA forms

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were completed and returned to the research team for scoring and interpretation within a few weeks of the other assessments.

Data Analysis

Descriptive statistics were used to report participant profiles on the measures. Summaries of group performance were reported, and the percentage of children below age expectations were calculated. Partial correlations, adjusting for age and IQ (see Licht, 1995), tested associations between sensory processing variables and occupational performance using the Statistical Package for the Social Science version 10.0 (SPSS, 1999).

Results

Demographic Correlates

Table 1 presents demographic information about the cognitive levels (standardized IQ scores) and ages of the children in our study as well as presents the intercorrelations between these demographics and the various measures used in the study. The scores on the VABS were positively correlated with IQ; SFA scores and Sensory Profile scores were positively correlated with age. Thus, these two demographic variables (age in months, IQ) were statistically controlled using partial correlations in the final analyses to remove potentially confounding influences of maturational factors and general cognitive ability on performance for those measures that were correlated (Licht, 1995).

Correlations Among Sensory Measures

Pearson correlation coefficients were calculated to see the relationships among the sensory processing measures used. We found that the TDDT-R external-control score and the Sensory Approach–Avoidance Rating were not related ($r = -.11$, $p > .05$); however, the TDDT-R internal-control score was correlated with the Sensory Approach–Avoidance Rating ($r = .62$, $p < .05$). The SSP was not correlated with either the TDDT-R (external-control score, $r = .29$, $p > .05$; internal-control score, $r = .20$, $p > .05$) or the Sensory Approach–Avoidance Rating ($r = .09$, $p > .05$), indicating that these measures perhaps were tapping different aspects of sensory processing. These findings are not surprising given that the Sensory Profile includes both hyporesponsiveness and hyperresponsiveness patterns, whereas the other measures represent only hyperresponsiveness patterns.

Table 1. Correlations Between All Measures and IQ and Age

Variable	IQ	Age
Sensory processing component		
Short Sensory Profile	.12	.65*
Sensory Profile–Group 1 items	–.07	.65*
TDDT-R (external score)	–.23	–.03
TDDT-R (internal score)	.23	.13
Sensory Approach–Avoidance Rating	–.49	.21
Occupational performance		
Self-care (VABS)		
Daily living skills	.59*	.18
School function (SFA)		
Part I: total participation	.06	.64*
Part III: total physical tasks	.19	.83**
Part III: total cognitive–behavioral tasks	.56	.78**
Play duration with novel toys	.44	–.01

Note. $n = 15$. SFA = School Function Assessment; TDDT-R = Tactile Defensiveness and Discrimination Test–Revised; VABS = Vineland Adaptive Behavior Scales.
*$p < .05$. **$p < .01$.

Descriptive Findings

Descriptive analyses provide details of the range of individual differences present in our sample. Several commonalities among the children were noted; however, not all were found to have significant sensory processing problems despite the fact that all 15 had full-mutation FXS. For example, on the SSP, the average score was more than 2 standard deviations below the mean of the typical reference sample (see Table 2). The majority of boys ($n = 11$) had scores that indeed fell more than 2 standard deviations below the norm. However, 2 obtained scores in the typical ranges, and 2 had scores that fell between 1 and 2 standard deviations from the norm.

Although reference norms are not available for children developing typically on either the TDDT-R or the Sensory Approach–Avoidance Rating, our observations from previous pilot work indicated that sensory defensive behaviors are quite unusual for school-aged children who are typically developing and without sensory modulation disorders; scores for children developing typically tend to show floor effects (total scores near 0) on both scales. In our FXS sample, we found that 2 boys had scores that fell at or near 0 on the TDDT-R; 3 showed low scores (few concerns) on the Sensory Approach–Avoidance Rating. The remainder had scores demonstrating varying levels of sensory difficulties, with 4 showing very high levels of aversion–avoidance on at least one of the two scales.

The range of performance across occupational domains of play, self-care, and school function also varied among the children, although both the SFA criterion scores and VABS standard scores were generally much below the range of typical performance in the reference samples. Reference scores are not available for the measure of play duration. On the VABS, the mean standard (composite) score was 56.4 ($SD = 13.5$), falling near the cut-off between the mildly to moderately delayed range of performance. Compared with the norms for samples of children who are typically developing, all but 1 boy fell more than 2 standard deviations below the

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Table 2. Descriptive Statistics for All Measures for Boys With Fragile X Syndrome

Variable	Range of Scores Possible	Fragile X Syndrome Sample: Range	*M*	*SD*
Sensory processing				
Short Sensory Profile	38–190	75–164	131 26	
Sensory Profile–Group 1 items	65–325	143–281	226	37
TDDT-R (total external score)	0–48	0–40	20	12
TDDT-R (total internal score)	0–33	0–14	7	4
Sensory Approach–Avoidance Rating	0–18	0–8	5	3
Occupational performance				
Self-care (VABS) *M* = 100 (*SD* = 15)[a]	28–71	56	14	
School function (SFA criterion scores)				
Part I: participation	0–100	45–100	73	20
Part III: sum physical tasks	0–1200	488–1062	840	174
Part III: sum cognitive–behavioral tasks	0–900	243–767	545	158
Play duration with novel toys (in sec)	0–1800	163–622	356	157

Note. $n = 15$. SFA = School Function Assessment; TDDT-R = Tactile Defensiveness and Discrimination Test–Revised; VABS = Vineland Adaptive Behavior Scales.
[a]The VABS is a standardized, norm-referenced test; thus, the exact mean and standard deviation is presented for the normative sample.

mean; 4 boys' scores fell more than 3 standard deviations below the mean.

We note that the SFA uses criterion scores that reflect the teacher's perceptions of the child's functioning levels compared with their same-aged peers in the same classroom; thus, the scores are not intended as a measure of normative performance. However, for descriptive purposes, full participation on the SFA Part I would be indicated by criterion scores near 100. In our sample, 3 boys received a full participation score of 100 on Part I of the SFA, and 1 additional boy received a score of 93, indicating that these 4 boys were participating in school functional activities at or near the level of their classroom peers. The remainder of the sample showed relatively low participation in school tasks compared with peers.

Correlational Findings: Sensory Processing and Occupational Performance Areas

Pearson partial correlations were used to examine relationships between each of the sensory processing variables and each of the occupational performance measures, controlling for effects of age and IQ for those measures that were correlated with these demographic variables. Both conventional ($p < .05$) and conservative ($p < .01$) significance levels are reported (see Table 3).

Sensory processing and daily living skills. Results revealed a significant negative relationship between the TDDT-R internal-control score and the VABS score ($r = -.63$, $p = .02$), suggesting that the boys who demonstrated a lower level of self-initiated approach (more aversive–avoidance reactions) with tactile media were less independent in daily living skills.

Sensory processing and school function. The Sensory Approach–Avoidance Rating was significantly negatively correlated with all three of the SFA variables, indicating that children who demonstrated more aversive–avoidance behaviors (less engagement) while exploring sensory toys had relatively lower scores in school function. A moderately strong, but nonsignificant correlation was noted for the Sensory Profile with SFA cognitive–behavioral tasks. Although we predicted negative correlations, we found positive correlations of moderate effect sizes (nonsignificant) between the TDDT-R external-control score and the three SFA measures.

Sensory processing and play. As expected, Sensory Approach–Avoidance Rating scores were significantly nega-

Table 3. Correlations Between Measures of Sensory Processing and Occupational Performance

	Sensory Processing				
	Sensory Profile		TDDT-R		
Occupational Performance	Short Form	Total Group 1	External	Internal	Sensory Approach–Avoidance Rating
Self-care (VABS)					
Daily living skills	.01	.09	.40	−.63*	−.39
School function (SFA)					
Part I: total participation	.25	.33	.53	−.34	−.76**
Part III: total physical tasks	.28	.29	.31	−.09	−.79**
Part III: total cognitive–behavioral tasks	.49	.55	.52	.12	−.86**
Play duration with novel toys	−.04	.07	.11	−.38	−.56*

Note. $n = 15$. SFA = School Function Assessment; TDDT-R = Tactile Defensiveness and Discrimination Test–Revised; VABS = Vineland Adaptive Behavior Scales.
*$p < .05$. **$p < .01$.

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tively correlated with play duration ($r = -.56$, $p = .05$)—the children who demonstrated more aversive–avoidance behaviors toward sensory properties of toys also spent less time engaging in play with novel toys. A moderately strong negative correlation (not significant) was noted for the TDDT-R internal-control score and play duration.

Discussion

Overall group findings indicated that these 15 boys with FXS were found to perform at ranges much below "typical" levels on sensory processing functions and "criterion" cutoffs for occupational performance relative to classroom peers. However, significant individual differences were noted despite the fact that all of the boys had the full-mutation FXS and significant developmental delays. Surprisingly, 3 boys (20%) performed within normal ranges on at least one of the measures of interest.

This study supports our hypotheses that controlling for age and IQ, some measures of sensory processing are associated with measures of occupational performance; however, these associations were not always in the direction predicted, and not all measures were equally associated. These empirical findings support existing clinical literature (e.g., Hagerman, 1996; Stackhouse, 1994) and further clarify the nature of individual differences.

Specifically, those children with greater *internally controlled* aversion (avoidance) to sensory features of toys, as measured by either the TDDT-R or the Sensory Approach–Avoidance Rating, had lower levels of participation and performance in school activities, were less independent in self-care at home, and engaged for shorter durations of play with novel toys. These avoidance responses appear to be a coping strategy used to deal with high levels of arousal, defensiveness, or fear of new sensory experiences, characteristics known to be prevalent in FXS (Belser & Sudhalter, 1995; Boccia & Roberts, 2000; Cohen, 1995). Dunn (1997) also described a pattern of "sensation avoiding" for persons attempting to counteract low neurological thresholds to stimulation. Many learning experiences inherently require approach and engagement in novel tasks; however, for some boys with FXS, low sensory tolerance may add challenges to their self-regulation and coping abilities and, ultimately, to their participation in daily occupations.

Our findings can be further elucidated through the lens of dynamical systems theory (Kamm, Thelen, & Jensen, 1990; Thelen, 1989), demonstrating how a developing system chooses the most stable, comfortable, or "efficient" pattern or solution given a specific context and developmental time point. For children with FXS, avoidance of sensory features may be one such stable pattern; however, if the pattern is not flexible enough to accommodate new challenges, exploration, adaptation, and occupational performance may be affected.

Considerable variability was evident across the children in our study, and intercorrelations were not uniformly strong across the measures. The Sensory Profile indicated that 11 of the 15 boys (73% of our sample) had definite sensory modulation differences, but this measure was not significantly associated with occupational performance. These scores, however, were significantly related to age such that the younger boys had greater levels of sensory symptoms than the older boys. Given that the Sensory Profile merges hyporesponsive and hyperresponsive patterns into one total score, this measure may obscure some potential relationships between sensory processing and occupational performance in a population prone to hyperresponsiveness. Use of the factor scores on the Sensory Profile may be indicated in future work.

Our results demonstrated outcomes in both positive and negative directions; these findings suggest that perhaps there is not a direct (causal) relationship between sensory processing vulnerabilities and occupational performance. Rather, these variables may have a dynamic interrelationship mediated by other factors producing an array of individual performance outcomes. That is, sensory processing components, however "deficient" in children with FXS, are likely not a sufficient explanation for difficulties in occupational performance. Deficits in participation likely arise from a transaction of multiple factors, including the context that may or may not support a child's intrinsic vulnerabilities.

As an example, we found that high levels of aversion to touch from unpredictable external sources (that the child cannot easily avoid) indicated a tendency toward increased independence in self-care and greater participation in school activities in some children with FXS studied. Although these results seemed potentially incongruent with our other findings and previous speculations in the occupational therapy literature (Kimball, 1993; Kinnealey et al., 1995), it appears that sensory processing vulnerabilities per se did not translate automatically into decreased occupational performance. The relationship between these variables appears more dynamic than linear, consistent with dynamical systems theory (Law et al., 1994). Furthermore, it is possible that in some cases, these sensory processing vulnerabilities may actually facilitate other adaptive coping or self-regulation strategies ("I'll do it myself") if, for example, protective factors (high intellectual abilities) or appropriate environmental supports (calm atmosphere) are available to an individual child. These findings speak to the importance of using dynamic, occupation-centered interventions that incorporate the child's strengths and facilitate

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effective coping strategies that mitigate a constrained sensory processing system, perhaps bypassing the need to remediate some "deficits" altogether.

Conclusion

These preliminary findings clarify some complex issues surrounding sensory processing in a special needs population. Independent of general maturational and cognitive factors, avoidance of self-controlled sensory experiences significantly correlated with decreased occupational performance in the areas of self-care, school function, and play in this sample of boys with FXS. However, tremendous individual differences were noted—some children with high levels of aversive responses to sensory experiences do quite well participating within supportive environments or in situations where they exert adaptive self-regulatory strategies, such as performing tasks more independently. These findings provide insight into understanding the dynamic nature of self-organizing systems and the variables potentially mediating the relationship between sensory processing components and complex occupational behaviors.

Replication studies with larger samples of children with FXS, as well as children with other developmental disorders, are needed to determine the generalizability of these findings. Further research also needs to address the limitations inherent in some measures developed for this study. For example, better-validated assessments of play across multiple contexts would be useful. Additionally, given that many of the significant findings depended on the validity of the Sensory Approach–Avoidance Rating, clinical interpretations warrant caution until further validation and replication studies are conducted. ▲

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