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Diagnostic Validity of Sensory Over-Responsivity: A Review of the Literature and Case Reports

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Abstract Atypical responses to sensory stimulation are frequently reported to co-occur with diagnoses such as autism, ADHD, and Fragile-X syndrome. It has also been suggested that children and adults may present with atypical sensory responses while failing to meet the criteria for other medical or psychological diagnoses. This may be particularly true for individuals with over-responsivity to sensation. This article reviews the literature related to sensory over-responsivity and presents three pediatric cases that present a profile of having sensory over-responsivity without a co-occurring diagnosis. Findings from these cases provide very preliminary evidence to support the suggestion that sensory over-responsivity can occur as a sole diagnosis. Within this small group, tactile over-responsivity was the most common and pervasive form of this condition.

Keywords Sensory processing · Sensitivity · Modulation · Sensory integration · Defensiveness

Diminished or exaggerated responses to typical sensory stimulation have been described as a feature of many prominent diagnostic conditions. The terms under-responsiveness and over-responsiveness have appeared as the most current descriptors of these behaviors (Miller et al. 2007). Attempts to understand, classify, research, and treat both under and over-responsiveness have been ongoing since they were first described by Ayres (1965). None-the-less, after 40 years of work, our understanding of sensory processing patterns within and across groups, and their relationship to functional behaviors, continues to be imprecise.

The umbrella term Sensory Modulation Dysfunction (SMD) is currently being used to encompass both over and under-responsivity, along with the overlapping or fluctuating responsivity. Sensory seeking has also been suggested as a subtype of SMD (Miller et al. 2007). This has been questioned, however, since seeking behaviors have been found in populations exhibiting both over and under-responsive (Liss et al. 2006). Liss and colleagues have suggested that sensory seeking is more a compensatory mechanism used to moderate high arousal levels, at least by children with Autism Spectrum Disorder.

Some investigators have attempted to identify subgroups of individuals with sensory processing disorder, with the most prominent model presented by Dunn (Dunn and Brown 1997; Dunn 1999). Dunn's model suggests that individuals could be classified based on their neurological threshold and behavioral responses to incoming stimuli. According to this model high neuronal thresholds are indicative of a nervous system that requires a stronger or more intense input to elicit a behavioral response, suggestive of an under-responsive nervous system. Lower neuronal thresholds, on the other hand, are indicative of a nervous system that requires less intense or less frequent stimulation to fire, suggestive of an over-responsive nervous system. The model further highlights that individuals can respond either in accordance with their neurological threshold, or act to counteract their threshold. For example, individuals with high thresholds acting in accordance with

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those thresholds may appear passive or non-responsive. Dunn classified these individuals as having low-registration. Individuals with high thresholds may also seek out additional sensory input to counteract that threshold; Dunn classified these individuals as sensory seeking. Similarly, individuals who have a low threshold and act in accordance with their threshold would be considered sensory sensitive. Yet individuals who act to position themselves away from potentially noxious stimuli would be classified as sensory-avoiders.

Models such as Dunn's provide a rich description of how neurological processes may influence personal tendencies to produce a range of observable behavioral responses. It is not surprising, therefore, that both neurological and behavioral measures have been used to investigate the broad characteristics of SMD; with neurological studies measuring reactions in specific brain structures or pathways, and behavioral measures focused on observable responses to sensation.

While looking at the entire scope of SMD may be beneficial for understanding the range of sensory responsiveness, there may also be a need to examine separately the features of sensory under and over-responsivity. Researchers attempting to identify sensory patterns in populations that include the full range of sensory responsiveness run the inherent risk of dampening findings due to the polar dimensions of under and over-responsivity. By considering under or over-responsiveness as separate aspects of the construct of SMD, researchers may be better able to understand the sensory-related behavioral patterns as a whole (Liss et al. 2006), and the essential links between neurological mechanisms and observable behavior. Physiological differences between children with over-responsivity and under-responsivity have been identified, providing preliminary evidence of behavioral–physiological correlations in SMD (McIntosh et al. 1999). Over-responsivity has been identified as the more common form of SMD (approximately 80%) based on referrals to research programs (Schaaf 2001). Therefore, this discussion and the case-studies presented will focus on the over-responsive form of SMD.

Behaviors associated with over-responsivity include actively avoiding stimuli as well as defensiveness or sensitivity to perceived unpleasant stimuli (Dunn 1999; Interdisciplinary Council on Developmental and Learning Disorders [ICDL] 2005; Lane 2002). Individuals with over-responsivity to sensation may withdraw from certain types of touch, cover their ears in response to everyday sounds, and/or avoid movement activities that are typically enjoyable or non-noxious to others. These individuals may also have limited diets due to sensitivity to the taste, smell or texture of certain foods. They may also get easily overwhelmed in certain environments, demonstrate strong emotional reactions to sensory stimuli, and engage in disruptive behaviors when demands become too great (Parham and Mailloux 2005).

Sensory over-responsivity has been identified in children and adults with specific diagnoses such as autism, Fragile X syndrome, attention deficit hyperactivity disorder (ADHD), and mood disorders (Baranek et al. 2002; Brown et al. 2002; Liss et al. 2006; Mangeot et al. 2001; Miller et al. 1999; Parush et al. 2007; Rogers and Ozonoff 2005). While it has been helpful to study sensory responsivity within these diagnostic categories, symptoms of over-responsivity have also been described in populations with no other co-existing conditions, suggesting a mechanism of over-responsivity with an independent etiology. This may be thought of in parallel to the symptom of inattention; a behavioral manifestation that spans multiple diagnoses but in its most intense form is acknowledged as a disorder (ADHD) that can occur in isolation. In contrast to ADHD, sensory over-responsivity has been only minimally described independent of other diagnostic groups (Kinnealey and Fuiek 1999; Kinnealey et al. 1995) and, secondarily, has failed to be unanimously recognized as an independent syndrome. In this paper we aim first to discuss the current means of measuring over-responsivity related to observable behaviors and neuro-physiological responses. A secondary aim is to highlight the occurrence of over-responsivity across recognized diagnostic categories. Finally we present three case studies we feel provide very preliminary support for the argument that over-responsivity can occur within an otherwise non-diagnostic population.

Measurement of Sensory Responsiveness

Electrophysiological Measurement

In typical individuals the ability to inhibit sensory responsiveness is considered an important trait. Measurement of the ability to inhibit sensory responsiveness has been accomplished electrophysiologically using two different paradigms related to sensory gating: P50 suppression and prepulse inhibition (PPI). Although both reflect sensory gating mechanisms, available evidence suggests that different neural circuits mediate the two responses (Hong et al. 2007). In addition to these gating paradigms, measurement of response to sensory stimuli has been conducted using electrodermal responses (EDR) (Blair 1999; Crowell et al. 2006; Hagerman et al. 2002; McIntosh et al. 1999; Miller et al. 1999; Venables 1977).

P50 Suppression

The P50 is an evoked response to sensory input, identifiable on an electroencephalogram (EEG). The wave is

present as a positive peak approximately 50 ms, and is the electrical response in the brain to a sensory stimulus. Evoked responses to sensory stimuli indicate that the specific pathway(s) of interest are intact and processing sensory input. In paradigms of P50 suppression two stimuli are presented in close succession; the response to the first stimulus (conditioning stimulus) induces suppression of the response to the second stimulus (test stimulus). This response is considered adaptive, a means by which the central nervous system avoids excessive activation. The typical inter-stimulus interval (ISI) for P50s is 500 ms. Inhibition of the second response is attributed to sensory gating, and it is measurable using event related potential (ERP). Although P50 suppression is often demonstrated using two auditory stimuli (intra-modal gating), it has also been demonstrated using visual and auditory stimuli (cross-modal gating) (Oranje et al. 2006).

Prepulse Inhibition (PPI)

PPI reportedly reflects an ability to buffer the central nervous system from what has been termed "the potentially chaotic flow of information and sensory stimuli" (Cadenhead et al. 1993, p. 1862). This stimulus gating paradigm uses paired stimuli, presenting a weaker first stimulus and a stronger second stimulus. The outcome measurement is motor startle response. The startle response, or reflex, is inhibited by the prepulse stimulus, and the degree of inhibition is a reflection of the degree of sensorimotor gating. Optimal inter-stimulus interval is reportedly 100 ms; cross-modal gating has not been demonstrated (Oranje et al. 2006).

Electrodermal Response (EDR)

Electrodermal responses (EDR) are changes in skin electrical conductance. They are the response of the eccrine sweat gland to a specific, phasic, stimulus (Andreassi 1989). EDR is not a reflection of gating. Instead this tool can be used to assess either strength of responsiveness or habituation to sensory stimuli. EDR to sensory stimuli have been shown to be abnormal in individuals with autism, schizophrenia, attention-deficit disorder, and conduct disorder (Blair 1999; Crowell et al. 2006; Hagerman et al. 2002; McIntosh et al. 1999; Miller et al. 1999; Venables 1977).

Other Physiological Measures

While the P50 wave is the most reported means of measuring sensory gating, other measures of EEG are being used to investigate neuro-physiological patterns related to sensory processing and sensory registration. For instance Parush and colleagues (2007) have measured somatosensory evoked potential (SEP) and Davies and Gavin (2007) used P50 to assess sensory responsivity along with P200 and N100 as measures of sensory registration. Additional physiological measures have also been used to look at nervous system responses to sensory challenges. Cardiac vagal tone has been presented as a means of investigating parasympathetic nervous system activity (Porges 1992, 1995). Initial results suggest that children with SMD do not recover as efficiently from sensory stressors as typical children (Schaaf et al. 2003). Salivary cortisol, a primary stress hormone, reflects stress response and recovery. Atypical levels of salivary cortisol may be indicative of maladaptive stress responses to benign sensory stimuli in populations of children with sensory over-responsivity (Reynolds 2006).

Behavioral Measurement of Sensory Responsiveness

Behaviorally, sensory modulation has been assessed primarily through the use of survey instruments such as the Sensory Profile (Dunn 1999), the Infant–Toddler Sensory Profile (Dunn 2002), and the Adolescent–Adult Sensory Profile (Brown and Dunn 2002). These tools use self or parent report to identify the frequency of behaviors in response to sensory stimuli including touch, vision, sound, taste, smell, and movement. Results from these assessments can be used to classify individuals as having sensory over-responsive and/or under-responsive behaviors. They can also identify individuals' dominant behavioral patterns in the areas of sensory seeking, sensory avoiding, sensory sensitivity, and/or low registration. Additional behavioral measures that have been introduced include the Sensory Experiences Questionnaire (SEQ) (Baranek et al. 2006), the Sensory Processing Measure (SPM) (Parham et al. 2006), the Sensory Over-Responsivity Scale (SensOR) (Schoen et al. 2005), and the Sensory Questionnaire (Liss et al. 2006).

Research within Diagnostic Groups

Sensory processing has traditionally been studied within diagnostic populations with a primary aim being the identification of patterns unique to individual diagnoses. This aim has been challenging to actualize due to the heterogeneity and co-morbidity inherent in diagnostic groups such as ADHD. The prevalence of SMD in diagnostic populations has been estimated at approximately 30%, though some investigators have suggested that this

Table 1 Summary of current research on sensory modulation in diagnostic groups

Author	Population characteristics	Sensory responsiveness characteristics				Identification tool*	Notes
		Under	NS**	Over	Typical		
Perry et al. (2007)	Adults with autism			x		PPI	Correlated with increased ratings of restricted and repetitive behaviors
Kemner et al. (2002)	Children with autism ages 7.3–13.6				x	P50 Gating	
Miller et al. (2001)	Children with autism	x				EDR	Physiological pattern of under-responsivity with contrasting behavioral scores more suggestive of over-responsivity
		x		x		SSP	
Leekam et al. (2006)	Children with Autism ages 2.8–11.1 years		x			DISCO	94% sensory abnormalities in children with autism compared to 33% typical and 65% with DD and Language Impairment
Baranek et al. (2006)	Children with autism 5–80 months	x		x		SEQ	69% overall sensory symptoms in children with autism; subset of children with both under and over-responsiveness found
Kern et al. (2006)	Individuals with autism ages 3–56 years	x		x		SP	Significant differences from controls even when over and under-responsive items examined separately
Talay-Ongan and Wood (2000)	Children with autism ages 4–14 years	x		x		SSQ-R	Sensory sensitivities were found to increase with age
Watling et al. (2000)	Children with autism ages 3–6 years	x		x		SP	Oral sensitivity only over-responsive category where differences were found
Tomchek and Dunn (2007)	Children with autism ages 3–6 years	x		x		SSP	95% of sample demonstrated sensory processing differences; greatest differences found in under-responsive/seeks sensation, auditory filtering and tactile sensitivity
O'Riordan and Passetti (2006)	Children with autism (mean age 8 years 7 months)			x	x	Discrimination tasks	Enhanced auditory discrimination in children with autism; typical tactile discrimination skills
Liss et al. (2006)	Individuals with autism spectrum disorders, mean age 102.4 months (SD = 50.1 months)	x		x		ExSP	Both over-reactivity and under-reactivity found in sampled population but clustered with distinct behavioral patterns. Sensory seeking behaviors found in both sub-groups
Jones et al. (2003)	Adults with HFA			x		Qualitative analysis of internet sites	
McAlonan et al. (2002)	Adults with Asperger Syndrome			x		PPI	Reduced PPI in Asperger's group compared with controls
Blakemore et al. (2006)	Adults with Asperger Syndrome			x	x	Controlled experiment	Adults with Asperger's evidenced lower tactile thresholds at 200 Hz but not at 30 Hz. Also rated tactile stimulus to be more intense than controls
Dunn et al. (2002)	Children with Asperger Syndrome ages 8–14 years	x		x		SP	Children with Asperger's differed on 22/23 items on the Sensory Profile compared to control group
Pfeiffer et al. (2005)	Children with Asperger Syndrome ages 6–17 years	x		x		SP or adolescent/adult SP	Significant relationship reported between anxiety and sensory over-responsivity. Also between depression and sensory under-responsivity
Rogers et al. (2003)	Children with: autism			x		SSP	Children with autism and Fragile X more impaired then developmentally disabled and typical group
	Fragile X	x		x			

Table 1 continued

Author	Population characteristics	Sensory responsiveness characteristics				Identification tool*	Notes
		Under	NS**	Over	Typical		
Miller et al. (1999)	Individuals with Fragile X mutation and Fragile X Syndrome ages 4–49 years			x		EDR	Individuals with Fragile X demonstrated significantly higher magnitudes, more responses per stimulation, and lower rates of inhibition indicating enhanced reaction to sensation
Roberts et al. (2006)	Boys 1–11 years with Fragile X			x		Vagal Tone	Lower baseline levels and less vagal reactivity than typical controls in response to a task demand
Frankland et al. (2004)	Boys with Fragile X			x		PPI	PPI impairments in the fragile X children predicted the severity of their IQ, attention, and adaptive behavior
Chen and Toth (2001)	Fragile X mice			x		PPI	
Baraneket al. (2002)	Boys with Fragile X ages 4–10			x		SP TDDT-R SA-AR	Boys who demonstrated more avoidance or aversive behaviors had lower scores on school function, independence in daily living skills and spent less time engaged in play with novel toys
Mangeot et al. (2001)	Children with ADHD ages 5–13 years	x		x x		SSP EDR	ADHD group had significantly higher scores in all domains of SSP and higher EDR peak magnitudes upon initial presentation of sensory stimuli
Parush et al. (2007)	Boys with ADHD 5–11 years of age			x		EEG	Boys with ADHD and tactile defensiveness had higher somatosensory evoked potential (SEP) amplitudes than boys with ADHD only or typical boys
Castellanos et al. (1996)	Boys with ADHD & Tourette's Syndrome			x	x	PPI	Reduced PPI in subjects with co-morbid ADHD and Tourette's syndrome not ADHD only
Yochman et al. (2004)	Boys with ADHD	x		x		SP	Differences found in general sensory processing, overall modulation, and behavioral and emotional responses
Dunn and Bennett (2002)	Children with ADHD	x		x		SP	Differences found on all sections of the Sensory profile
Kalpogianni (2002)	Children with ADHD	x				SP	Differences found in areas of sensory seeking, emotional reactivity, and inattention/distractibility
Olincy and Martin (2005)	Adults with BPD			x		P50	Subjects had history of at least one manic episode
Rich et al. (2005)	Children with BPD				x	PPI	BPD subjects were medicated and not acutely manic. No differences in PPI or habituation from typical controls
Kruger et al. (2006)	Adults with BPD			x		EEG	Low odor thresholds in euthymic patients with event-triggered mood episodes
Lyoo et al. (2006)	Adults with BPD		x			Brain MRI	Cortical thinning in sensory and sensory association cortices
Brown et al. (2002)	Adults with: BPD Schizophrenia	 x		x x		Adult SP	BPD and Schizophrenic subjects had higher levels of sensory avoiding than controls. Schizophrenic group also higher on low registration and sensory seeking
Perry et al. (2001)	Adults with: BPD Schizophrenia			x x		PPI	No significant difference between BPD and schizophrenic subjects. Significantly lower PPI than control subjects

Table 1 continued

Author	Population characteristics	Sensory responsiveness characteristics				Identification tool*	Notes
		Under	NS**	Over	Typical		
Cadenhead et al. (2000)	Adults with Schizotypal personality disorder			x		P50	Subjects with schizotypal personality disorder had significantly less P50 suppression than normal subjects

*SP, sensory profile; SSP, short sensory profile; SEQ, sensory experiences questionnaire; DISCO, diagnostic interview for social and communication disorders; PPI, pre-pulse inhibition; P50, P50 sensory gating; EEG, electroencephalogram; EDR, electro-dermal reactivity; MRI, magnetic resonance imaging; TDDT-R, tactile defensiveness and discrimination test-revised; SA-AR, sensory approach-avoidance rating; SSQ-R, sensory sensitivities questionnaire-revised; ExSP, expanded sensory profile

**NS, not specified. Results presented in the article identified a sensory processing dysfunction without specifying over or under-responsivity

number is closer to 100% in children with autism (Baranek et al. 1997; Leekam et al. 2006; Tomchek and Dunn 2007). These estimates do not, however, discriminate between over and under-responsiveness and are based primarily on behavioral, not physiological, measures. Table 1 highlights the current research related to sensory modulation in six diagnostic populations: ADHD, autism, Asperger's syndrome, bipolar disorder, schizophrenia, and Fragile X syndrome.

Autism is the most high profile diagnosis in which differences in sensory responsiveness have been identified. General patterns of sensory processing have been widely examined in both children and adults on the autism spectrum using a variety of methods. Both over and under-responsive behaviors, sometimes in combination, have been reported in persons with autism and Asperger's syndrome using sensory-based questionnaires (Baranek et al. 2006; Dunn et al. 2002; Kern et al. 2006; Leekam et al. 2006; Liss et al. 2006; Miller et al. 2001; Pfeiffer et al. 2005; Talay-Ongan and Wood 2000; Watling et al. 2000). Physiologically, a reduced PPI has been noted in adults with autism (Perry et al. 2007) and also adults with Asperger's syndrome (McAlonan et al. 2002). In children with autism, however, EDR data suggests a pattern of under-responsiveness (Miller et al. 2001) while P50 gating abilities are reportedly similar to those of typical controls (Kemner et al. 2002).

While generally thought to be sensation-seekers, children with ADHD have been reported to have difficulties in all areas of sensory processing and sensory modulation. Parush and colleagues (2007) found that 69% of boys with ADHD referred to their study also demonstrated tactile defensiveness. EEG recordings from this study indicated that those children with ADHD + tactile defensiveness had higher somatosensory evoked potentials than children with ADHD only or typical children. Children with ADHD have also evidenced significantly higher EDR magnitudes upon initial presentation of sensory stimuli during a sensory challenge (Manegot et al. 2001). Contrary to these findings, PPI was found to be normal in a small sample of boys with ADHD only, but reduced in boys with ADHD and Tourettes Syndrome (Castellanos et al. 1996).

Two diagnostic groups in which sensory processing has been largely studied from a physiological rather than a behavioral perspective are bipolar disorder (BD) and schizophrenia. These two diagnoses were identified as separate and distinct conditions around the late 19th century but continue to share commonalities in prevalence, age of onset, familial aggregation, and heritability (Maier et al. 2006).

Behavioral data using the Sensory Profile suggests that individuals with BD have a higher rate of sensation avoidance (suggestive of sensory over-responsivity) than typical controls. For adults with BD, physiological data using EEG, PPI, and P50 sensory gating supports the behavioral evidence that they may in fact have over-responsiveness to sensory stimuli (Kruger et al. 2006; Olincy and Martin 2005; Perry et al. 2001). Some researchers have questioned the existence of a purely BD population, however, due to high rates of co-morbidity ranging from 44 to 100% with conditions such as ADHD, anxiety disorder, conduct disorder, and oppositional defiant disorder (Caetano et al. 2005).

Subjects with schizophrenia have also showed significant differences in the area of sensation avoidance on the Sensory Profile. However, they also scored outside of typical range on dimensions of low registration and sensation seeking, suggesting a mixed profile of under and over-responsivity (Brown et al. 2002; Waltermire et al. 2007). Support for sensory over-responsive behaviors in individuals with schizophrenia have been found using PPI and P50 sensory gating (Cadenhead et al. 2000; Perry et al. 2001).

Lastly, in individuals with Fragile X, behavioral measures have indicated a consistent pattern of over-responsiveness to sensory stimulation (Baranek et al. 2002; Rogers et al. 2003). This is supported by physiological measures of elevated EDR magnitudes (Miller et al. 1999),

reduced cardiac vagal tone (Roberts et al. 2006), and suppressed PPI (Frankland et al. 2004).

Thus, while conflicting data do exist, there is evidence to suggest that many individuals in these diagnostic groups experience sensory over-responsiveness. This has likely fueled the argument that differences in sensory responsiveness are solely features of other diagnostic issues rather than an independent diagnosis. However, early evidence suggests that SMD is a stand-alone diagnosis for some individuals.

Research on Populations with SMD-only

Sensory over-responsivity, in the form of tactile defensiveness, was first identified by Ayres (1965) as a type of sensory integration dysfunction. Ayres proposed a theory of sensory integration as a means of explaining the underlying cause of sensorimotor and learning deficits in children (Ayres 1972a, b). She defined "sensory integration" as the neurological processes used to organize sensation from the body and the environment, leading to effective environmental interactions (Ayres 1972b). Deficits in sensory integration would be reflected in behavioral, social, academic, or motor coordination problems. Such deficits encompassed sensory modulation disorders as well as disorder of sensory discrimination and praxis (Ayres 1972b). Ayres' published testing materials focused on the aspects of sensory integration that were reliably and objectively measured, such as praxis and sensory discrimination (Ayres 1972c, 1989). Sensory modulation deficits, such as tactile defensiveness, were identified through structured clinical observation (Ayres 1969).

Current research with subject populations comprised of individuals with SMD has been questioned since SMD is not yet recognized as an ICD-10 or DSM-IV diagnosis (Cheng and Boggett-Carsjens 2005). However, the Diagnostic Classification of Mental Health and Developmental Disorders of Infancy and Early Childhood: Revised Edition (DC:0-3R) (Zero to Three 2005) and the Diagnostic Manual for Infancy and Early Childhood (ICDL-DMIC) (ICDL 2005) have both included Regulatory Sensory Processing Disorders as a diagnostic category, with over-responsive sensory modulation identified as a specific type of SMD associated with the fearful/cautious or anxious behavior pattern and the negative/defiant or stubborn behavior pattern.

Validity for the existence of SMD has been enhanced by measurement of physiological responses associated with sensory stimulation. For instance, McIntosh and colleagues (1999) studied EDR patterns in 19 children clinically diagnosed with SMD and without conditions such as cerebral palsy, fetal alcohol syndrome, or autism. Children in the experimental group showed behaviors consistent with SMD which were confirmed by parent interviews. Nineteen healthy controls were matched to the SMD group based on age and sex. Results showed that four of the children with SMD showed no response to stimulation suggesting a distinct under-responsive pattern. Excluding these four non-responders, the children with SMD showed larger and more frequent EDR than typical controls and slower habituation to repeat stimuli, suggesting an over-responsive pattern in approximately 79% of the experimental sample.

As is the trend in other diagnostic populations, emerging research related to SMD has included studies examining potential genetic factors and heritability traits. Goldsmith et al. (2006) conducted a population-based twin study to examine tactile and auditory defensiveness in young children. Results indicated moderate genetic influences with tactile defensiveness showing greater heritability. Both auditory and tactile defensiveness were correlated with fearful temperament (r = .13–.50) and anxiety (r = .21–.28) ($p < .001$).

Kinnealey and Fuiek (1999) also found anxiety levels to be elevated in a population of adults with over-responsivity and free from other psychopathology. Sensory over-responsivity was assessed using the ADULT-SI and the presence of psychopathology was ruled out using the Counseling Evaluation Test, a self-administered true–false questionnaire. Higher levels of self-reported anxiety and depression were found in the over-responsive vs. the non over-responsive adult populations. Kinnealey and colleagues (Kinnealey et al. 1995) have also documented sensory defensiveness in five adults in a phenomenolgic study. All adults presented without a history of physical or sexual abuse or hospitalization for emotional or psychological diagnoses. They reported lifetime experiences of sensory defensiveness in one or more sensory systems with tactile defensiveness occurring in all five subjects impacting self-care, choice of activities and patterns of intimacy. No such study has presented case-reports on children with over-responsivity without co-morbid diagnoses.

Case Study Reports

Research Methods

The purpose of the following case study reports was to identify and describe a set of children who exhibited behaviors of sensory over-responsivity and had no other co-existing neurological or psychological diagnoses. Through a process of parent questionnaires, parent interview, formal assessment and child observation the researchers aimed to confirm the presence of sensory over-

responsivity and the absence of any other conditions. Five children were referred for the study; three met the criteria to be included.

Sample

Potential participants were recruited through three local occupational therapists who were considered to be master-level clinicians. All referring clinicians had certification in the administration and interpretation of the Sensory Integration and Praxis Test and had at least 5 years of clinical experience. Potential "cases" were between the ages of 6 and 12, and identified by the master clinician as evidencing signs of sensory over-responsivity, and *without* any of the following medical or psychological diagnoses: ADHD, autism, anxiety disorder, seizure disorder, obsessive compulsive disorder, or any other neurological, genetic or mood disorders. The study was explained and permission to contact parents was obtained. The primary researcher contacted interested families by telephone, explained the study, and acquired parental consent and child assent.

Confirmation of Sensory Over-responsivity

Confirmation for the presence of sensory over-responsivity was established in two ways. First parents completed the Sensory Over-responsivity Inventory (SensOR), a 76-item questionnaire that assesses individuals' responses to various sensory stimuli in the domains of Tactile Sensitivity, Taste Sensitivity, Smell Sensitivity, Visual Sensitivity, Auditory Sensitivity, and Movement Sensitivity (Schoen et al. 2005). The SensOR was mailed to the parents along with the consent form and was returned to the researchers in the same pre-stamped envelope. Children whose behaviors ranked at least two standard deviations above the normative population in one of the six categories met the criteria for inclusion.

Sensory over-responsivity was also confirmed through parent interview. A confirmatory interview was conducted so that parents would have an opportunity to expand on behaviors that were not questioned on the SensOR, and also confirm that they felt that their child demonstrated sensory over-responsiveness. Interviews took place at a time and location that was convenient to the parent. Two interviews were conducted in the family's home and one was conducted in an empty waiting room at an occupational therapy clinic. In addition to being asked about their child's sensory behaviors, parents were asked whether or not their child had attended regular visits with their primary care physician. It was assumed that significant genetic conditions (i.e., Down's syndrome, Fragile-X syndrome, Rett's syndrome) or neurological disorders (i.e., cerebral palsy, Spina bifida, or seizure disorders) would have been identified if the child had attended regular visits with their primary care provider. If both the SensOR and parent interview corroborated with the master clinician's referral, and the child had attended regular visits to the primary care physician without being diagnosed with any medical conditions, the child was included as a case.

Ruling Out Potential Co-occurring Diagnoses

The Wechsler Abbreviated Scale of Intelligence (WASI) two-subtest scale was used to ensure that the all children presented with intelligence quotients above 80 to rule out the presence of intellectual disability. The two-subtest form of the WASI, comprising the Vocabulary and Matrix Reasoning subtests, was designed to provide a quick and accurate full scale IQ score. Authors of the WASI note that the two-subtest scale is sufficient for providing a general summary of an individual's cognitive functioning (Psych-Corp 1999). The full scale IQ for the two-subtest WASI correlates highly with the Wechsler Intelligence Scale for Children, Third Edition (WISC-III) ($r = .81$), and the Wechsler Adult Intelligence Scale, Third Edition (WASI-III) ($r = .87$). In two cases, the WASI was conducted in the child's home following the parent interview. One WASI was conducted in an occupational therapy clinic. Although the WASI is generally administered by psychologists, the first author received permission from the publisher to use the test under the supervision of a licensed Psychiatrist. The supervising psychiatrist was available for consultation and to review final scores.

The following two assessment tools were used to rule out the presence of additional diagnoses typically associated with sensory processing disorders.

The Childhood Autism Rating Scale (CARS) (Schopler et al. 1988) was used to rule out the presence of autism in these children. The CARS, a 15 item behavioral measure, was completed by the primary investigator who spent time observing and interacting with the child for a minimum of 1 h in their home and/or clinic setting. Using a 7-point scale the investigator indicated the degree to which the child's behavior deviates from that of a normative population. Norms for the CARS are based on a population of 1,500 children. Total scores on the CARS have been strongly linked to DSM-IV diagnostic criteria for autism and the CARS has been shown to have a high degree of sensitivity (.94) and specificity (.85) (Perry et al. 2005; Rellini et al. 2004). Scores on the CARS range from 15 (no signs of autism) to 60. Children who score between 30 and 60 are categorized as having autism on a range of mild to severe.

The Child Symptom Inventory-4 (CSI-4) (Gadow and Sprafkin 2002) was used to rule out the presence of additional psychological diagnoses. The CSI-4 includes two rating scales; one completed by the parent and the other by the child's teacher. The tool is designed to identify symptoms of common childhood psychiatric disorders including ADHD, oppositional defiant disorder, conduct disorder, separation anxiety, generalized anxiety, social phobia, specific phobia, depression, dysthymia, Asperger's syndrome, pervasive developmental disorder, schizophrenia, obsessive compulsive disorder, posttraumatic stress disorder, motor tic disorder, and vocal tic disorder. The parent checklist contains 97 items covering 17 disorders, while the teacher checklist contains 77 items related to 13 disorders. The test provides normative scores and clinical cut off scores for children between the ages of 5 and 12. Test–retest scores reported for the CSI-4 range from .61 to .88 (satisfactory) with a high degree of internal consistency (.72–.94) (Mattison et al. 2003). The CSI-4 parent form was completed at the time of the parent interview. Parents were responsible for giving the CSI-4 form to the child's teacher who mailed the pre-coded form directly to the researchers in a pre-stamped envelope. All forms were completed and returned to the researchers.

Scores for standardized questionnaires were obtained and compared to standard norms to determine whether or not the child's scores fall within typical range. As stated previously, five children were referred for this study and three met criteria to be included as a case. One child was excluded because he did not score as having sensory over-responsive behaviors on the SensOR. The second child was excluded because CSI-4 scores indicated potential psychological diagnoses including anxiety disorder and social phobia.

Case Study 1

Cody is an 11-year-old Caucasian male enrolled in a regular fifth grade classroom and receiving private occupational therapy services. He was referred to occupational therapy at age 10 due to concerns with fine motor and self-care skills. According to parent report he had been tested at school but did not qualify for the gifted program nor did they find any signs of learning disabilities. Thus, Cody did not qualify for any services in the school. Cody's birth history was unremarkable for pre-maturity, jaundice, or other complications. He reportedly met all early developmental milestones. However, parents noted that Cody had been a fussy baby and would refuse the bottle unless it was heated within a specific temperature range.

On the SensOR Cody scored outside of two standard deviations in the categories of Tactile Sensitivity and Taste Sensitivity. During the parent interview it was reported that difficulties at home were primarily around self-care activities. Mom noted that Cody does not comb his hair; describing the comb itself as a "mace-laser." Cody also described a wash cloth (used for washing his face) as a "sand-blaster." He dislikes nail clipping, hair washing, and tooth brushing. Cody also reported that he does not like to wear rough clothing such as jeans or stiff shirts. In the area of taste sensitivity, parents reported that Cody prefers tough–rough foods such as beef jerky and hamburgers. He is hesitant with new or unfamiliar foods and generally will not eat foods that he considers to be lumpy, soft, or slimy.

Based on the screening done for this study, Cody's IQ was in the high-average range with a slightly higher score in the verbal vs. the performance category. On the CARS Cody was noted to have low muscle tone, mild incoordination, and mildly abnormal responses to taste, smell and touch. Cody received a total score of 17 on the CARS, placing him in the non-autistic category. On both the CSI-4 parent and teacher forms, no diagnostic scores exceeded the screening cut-off point which would indicate the presence of a psychological disorder. In a space left for additional comments, Cody's teacher noted that Cody evidences difficulty controlling the volume of his voice, falls down often during recess, is awkward in his motor skills, and will often draw attention to himself in a negative way such as saying out loud, "I like being weird." Cody had attended regular physician visits and had no medical or psychological diagnoses.

Based on the tools used for this case-report, Cody presented with sensory over-responsivity in the areas of tactile and taste sensitivity. He scored within the high average range for cognitive abilities and showed no signs of autism or any other psychological diagnoses. Parents report regular trips to the family pediatrician who has given no indication that Cody has any genetic or medical conditions. Cody had never been taken to a neurologist or psychiatrist.

Case Study 2

Dominic is an 8-year-old Caucasian male twin born with strabismic amblyopia. Dominic's mother, a pediatric occupational therapist, identified some atypical behaviors when Dominic was around 18 months old. She described him as a disorganized child who had a difficult time at family parties and tended to melt down easily. He also had chronic ear infections as a child and continues to have allergies. Dominic received early intervention services but was no longer receiving occupational therapy at the time of this report. He was in the third grade and his parents noted that his biggest challenge was handwriting. At the time of data collection Dominic was also receiving

speech-language services for an articulation disorder. He was not diagnosed with any learning disabilities and visual impairments, present at birth, were resolved through the use of corrective lenses.

On the SensOR, Dominic scored outside of two standard deviations in the categories of Tactile Sensitivity and Auditory Sensitivity. Parents reported that the biggest sensory-related challenges at home were due to tactile sensitivities, especially dressing. Dominic strongly disliked seams, tags, or elastic in clothing; and especially disliked wearing socks or underwear. He also disliked having his nails clipped, having his hair cut, and brushing his teeth. Other tactile sensitivities included disliking the feel of glue, paint, hair-care products, kissing, and light stroking touch. In the auditory domain it was noted that Dominic disliked the sound of appliances (e.g., blender, vacuum, and hair dryer) and the sounds associated with large gatherings. Also, Dominic's parents also noted that he is bothered by the radio, TV, or someone talking when he is trying to concentrate. Dominic had attended regular physician visits and had no medical or psychological diagnoses.

Based on the screening done for this study, Dominic's IQ was in the average range with commensurate scores in the verbal and performance categories. On the CARS Dominic was noted to have mild incoordination, mild abnormal listening responses, and mild abnormal responses to taste, smell and touch. Dominic received a total score of 17.5 on the CARS, placing him in the non-autistic category. On both the CSI-4 parent and teacher forms, no diagnostic scores exceeded the screening cut-off point which would indicate the presence of a psychological disorder. In a space left for additional comments, Dominic's teacher noted that he sometimes has difficulty when attending assemblies or at lunch in the loud cafeteria but is able to read all test material on his own as long as the environment is quiet.

Based on the tools used for this case-report, Dominic presented with sensory over-responsivity in the areas of tactile and auditory sensitivity. He scored within average range for cognitive abilities and showed no signs of autism or any other psychological diagnoses. Parents reported regular trips to the family pediatrician who has given no indication that Dominic has any genetic or medical conditions aside from allergies.

Case Study 3

Sarah is a 12-year-old Caucasian female enrolled in a regular sixth grade classroom and receiving private occupational therapy services. Sarah had been referred to occupational therapy 2 months prior to enrollment in this study due to parent concerns regarding Sarah's personal hygiene and intolerance for wearing a bra. According to parent report, Sarah was born 1 week early but had no illnesses or birth complications. Parents noted, however, that Sarah had night terrors until about the age of four and did not like the movement of her child swing. By kindergarten, Sarah had major tantrums related to tooth brushing and dressing. Mom noted that she would sometimes have to bring Sarah to the guidance counselor's office in elementary school dressed in her pajamas and carrying her school clothes. At the age of five Sarah began counseling with a licensed social worker but continued to have a limited repertoire of clothes she would wear. Sarah maintained a restricted diet and became very upset when prompted to try new foods.

On the SensOR, Sarah scored outside of two standard deviations in the categories of Tactile, Taste, Smell, and Visual sensitivity. During the parent interview it was reported that difficulties at home continued to be centered on dressing, eating, and self-regulating in environments such as the bowling alley or mall. Sarah continues to have a limited diet consisting primarily of chicken nuggets, hamburgers, potatoes and fried foods. She will eat only one type of fruit (pears) and will not eat vegetables.

Based on the screening done for this study, Sarah's IQ was in the very superior range. On the CARS mild abnormalities were noted in Sarah's ability generate appropriate emotional responses, use her body in a coordinated manner, adapt to change, and respond appropriately to sound, taste, and touch. Sarah received a total score of 21 on the CARS, placing her in the non-autistic category. On both the CSI-4 parent and teacher forms, no diagnostic scores exceeded the screening cut-off point which would indicate the presence of a psychological disorder. Teachers noted that Sarah was creative and bright. Sarah had attended regular physician visits and had no medical or psychological diagnoses.

Based on the tools used for this case-report, Sarah presented with sensory over-responsivity in the areas of tactile, visual, gustatory, and smell sensitivity. She scored within the very superior range for cognitive abilities and showed no signs of autism or any other psychological diagnoses. Parents report regular trips to the family pediatrician who has given no indication that Sarah has any genetic or medical conditions.

Discussion

Case Study Reports

Taken together these three case studies provide very preliminary support for the existence of sensory over-responsiveness in the absence of other diagnostic

conditions. While it is clear that a diagnostic grouping cannot be based on three case reports, these reports do provide an initial foundation upon which sensory over-responsivity as a unique diagnostic entity can be considered. The research methodology used in this study may also be used as a starting point for additional prospective studies or retrospective reviews on the prevalence of SMD in non-diagnostic populations.

In looking at the three cases presented, the unifying link appears to be over-responsivity to tactile stimulation. It is both the most consistent area of deficit identified on standardized tools, and the major issue identified by parents as interfering with family routines and activities of daily living. This finding was consistent across the age range represented by three children in this study, and was identified in both girls and boys. This is interesting since tactile defensiveness was the first over-responsiveness behavior identified by Ayres (1964, 1965). It is also the one sensory system in which over-responsivity appears not to diminish with age (Kern et al. 2006).

Another interesting symptom observed across the three cases was motor incoordination. It is generally accepted that optimal motor output is guided by accurate and efficient intake and processing of sensory input (Pinel 2006). Individuals who have difficulty modulating sensory input, therefore, may be more likely to have less effective or efficient motor patterns. Another consideration is that children with both over-responsiveness *and* avoidance of sensory experiences may miss opportunities to engage in age-expected fine motor and gross motor tasks. It is unclear whether sensory-modulation and sensorimotor problems are distinguishable yet overlapping conditions, or if there is a potential causal or predictive relationship between the two constructs. It is also unclear whether sensorimotor problems would be present in children with other types of SMD, or whether this problem is unique to sensory over-responsivity. Future research should continue to explore these relationships.

Under-responsiveness and Sensory Seeking: Behavioral Considerations

While this study focused on one dimension of SMD, over-responsivity, there is a need to further examine the prevalence and behavioral manifestations of under-responsivity and sensory seeking. Evidence for the independent existence of sensory under-responsivity may be challenging to identify. Currently children falling into this category may be seen as having poor sensory registration (Dunn 1999), characterized by low energy levels and apparent disinterest in their surroundings. In both the DC:0-3 and the ICDL-DMIC under-responsiveness is reflected in children who are generally quiet and passive (ICDL 2005; Zero to Three 2005). In a population of children with autism, Liss and colleagues (2006) identified a cluster with under-responsivity that presented with the low adaptive functioning, communication impairments and deficits in social competence. This group was the lowest functioning overall of four sub-groups leading the authors to suggest that, in the autism population, under-responsivity may be related to mental retardation. A better understanding of the prevalence of under-responsivity in an otherwise typically developing population would help to clarify the mechanisms behind this sub-type of SPD and possibly help understand the correlations between under-responsivity, intellectual functioning, and adaptive behaviors.

According to Dunn's model, children with under-responsiveness to sensory input may also exhibit sensory seeking behaviors (Dunn and Brown 1997). These behaviors include a high level of activity and continuous engagement with the environment, in an attempt to glean all possible sensory input. It is uncertain whether sensory seeking is in fact a unique sub-category of SMD, with a distinct neurological pathology, or rather a behavioral manifestation observed in individuals attempting to alter or modify their levels of arousal. Liss and colleagues (2006) identified sensory seeking behaviors in both over-reactive and under-reactive children with autism. These authors suggested that children who are over-responsive may seek out pleasurable sensory activities, possibly as a means of escaping or avoiding more disturbing input. This challenges Dunn's conceptualization of sensory seeking as a means of counteracting a low neuronal threshold. Again, a better understanding of sensory-seeking in non-diagnostic populations may help us understand the inherent nature of the sensory modulation difficulties in the absence of other confounding factors related to diagnostic co-morbidities.

Directions for Future Research

Future studies using physiological measurements will help to substantiate sensory over-responsivity, and potentially sensory under-responsivity and sensory-seeking, as independent diagnostic categories, and to clarify the neurological mechanisms underlying the observable behaviors. In considering the design of future research studies with children with SMD, it will be pertinent to not only consider over- under-responsiveness in global terms, but also to examine sensory processing within and across specific sensory systems.

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