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Phenotypes within sensory modulation dysfunction

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Abstract

Sensory modulation disorder (SMD) is a severe inability to regulate responses to everyday sensory stimulation to which most people easily adapt. It is estimated to affect 5% to 16% of the general population of children. Although heterogeneity is seen in the presentation clinically, previous research has not empirically investigated whether the clinical heterogeneity of SMD can be classified into subtypes. This study explores a cohort of 98 children identified with SMD at the Department of Pediatric Rehabilitation by a member of the occupational therapy team at The Children's Hospital of Denver. Two subtypes of SMD were identified through cluster analysis based on data from 4 parent-report instruments. The first subtype is characterized by sensory seeking/craving, hyperactive, impulsive, externalizing (eg, delinquent, aggressive), unsocial, inadaptive, and impaired cognitive/social behavior. The second subtype is characterized by movement sensitivity, emotionally withdrawal, and low energy/weak behavior. Findings from this study present a step toward understanding and classifying the complexities of children with SMDs.

1. Introduction

Occupational Therapists have studied sensory processing since the early 1960s when Dr A. Jean Ayres [1-3] wrote the first scholarly articles in the field. However, questions remain about the validity of the diagnosis of sensory processing disorder (SPD), formerly referred to as *sensory integration dysfunction* by Ayres [3] and others [4]. Recently, a new diagnostic nosology was postulated to aid researchers in selecting homogenous samples and ultimately furthering the specificity of discussion related to theory, diagnosis, and intervention of sensory processing difficulties [5]. The new nosology differentiates 3 patterns of SPD: sensory modulation disorder (SMD), sensory discrimination disorder, and sensory-based motor disorder with subtypes noted within each pattern.

This study explores the diagnostic specificity within 1 of the 3 major patterns, SMD. Sensory modulation is the ability to regulate and organize the intensity and nature of responses to sensory input so that responses can be appropriately graded to the constantly changing sensory experiences of daily life. Sensory modulation disorder results in difficulty achieving and maintaining a developmentally appropriate range of emotional, attentional, and motoric responses to sensory stimuli [6-8], resulting in difficulty adapting to challenges encountered in daily life [8].

The clinical presentation of SMD varies with considerable heterogeneity in symptomatology [9]. One or more of the 7 sensory systems may be involved: tactile, vestibular, proprioceptive, visual, auditory, olfactory, and/or gustatory. Symptomatology includes sensory overresponsivity, sensory underresponsivity, sensory seeking/craving, or a combination of symptoms from the 3 subtypes. Atypical behaviors that result from SMD can range from severe to mild. Thus,

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clinical heterogeneity in SMD occurs in severity, number, manner, and which sensory systems are involved [4,5,7,10]. Given this heterogeneity, empirical evaluation of whether symptoms can be classified into subtypes is crucial for both assessment and intervention. Clinical diagnostic specificity related to types of SMD permits clinicians to better define and describe the child's sensory processing needs and guides appropriate intervention strategies.

Recent taxonomic efforts of SMD-type behaviors include the diagnostic manuals of 0 to 3 organization (DC:0-3) [11], which proposed 3 subtypes of SPD of regulation, and the Interdisciplinary Council of Developmental and Learning Disorders, which also proposed 3 subtypes of regulatory SPD [4].

The categories of SMD proposed by the diagnostic manuals of 0 to 3 organization and Interdisciplinary Council of Developmental and Learning Disorders were synthesized into 3 groupings within SMD by a recently proposed nosology [5]:

1. Sensory overresponsivity: a greater than typical response to sensory stimuli; responses to sensations are more intense, quicker in onset or longer lasting than those typically observed; the individual exhibits "fight, flight, or freeze" behaviors to sensation, for example, impulsive, aggressive, or withdrawn reactions.
2. Sensory underresponsivity*:* a disregard or passive response to sensory stimuli; responses are less intense or slower in onset than those typically observed; the individual is difficult to engage, lethargic, self-absorbed, and seems unaware of sensation, lacking an inner drive to explore sensory materials and environments.
3. Sensory seeking/craving*:* an intense, insatiable desire for sensory input; input is less than needed for the individual to feel satiated; individuals energetically engage in actions geared to adding more intense sensation, constantly moving, touching, watching moving objects, and/or seeking loud sounds or unusual olfactory or gustatory experiences.

Although these proposed patterns of SMD are clinically useful, there are limited empirical data related to the accuracy of the clinical categorization schemes that have previously been proposed for SMD. Ayres' original work [3] suggested one SMD pattern characterized by difficulty modulating tactile input, which she labeled tactile defensiveness. Later, Dunn [12,13] conducted a factor analysis of behaviors from her parent-report measure, the Sensory Profile, and proposed a quadrant classification scheme accounting for high versus low neurologic thresholds to sensory stimuli in combination with either a either passive versus active regulatory strategies. The 4 categories she proposed were sensation seeking, low registration, sensory avoiding, and sensory sensitivity. Miller et al [14] discussed a more complex model with multiple subtypes based on their ecologic model of sensory modulation that accounts for both internal and external factors affecting the ability to achieve homeostasis.

These models have face validity; however, empirical research defining subtypes is needed to clarify the variability in children with SMD and to determine if natural boundaries exist between subtypes. This clarification of subtypes will improve selection of homogenous samples in all applied SMD research, improve treatment planning for specific clinical cases, and decrease sample variation, thus increasing power in effectiveness research.

Many developmental and behavioral disorders are diagnosed based on the presence of a cluster of symptoms, for example, attention-deficit/hyperactivity disorder (ADHD), anxiety disorders, and autism spectrum disorders. Cluster analysis is a statistical technique commonly used to empirically define diagnostic taxonomies of complex disorders. Cluster analysis has been used to define subtypes of depression [15], to categorize suicidal patients into 3 subtypes [16], and to identify subtypes of eating disorders [17]. More recently, cluster analysis has been used to decipher subtypes and patterns in ADHD [18-20], bipolar disorder and other manic diseases [21-23], and the full spectrum of psychopathologic symptoms [24].

Cluster analysis is appropriate as an exploratory tool to define structure within data and reflect more homogenous patterns within groups and more diverse patterns across groups. This study uses cluster analysis to group behaviors in children with SMD based on the hypothesis that SMD clusters into meaningful subtypes (sensory overresponsivity, sensory underresponsivity, and sensory seeking/craving) based on behavioral characteristics of attention, sensation, and emotion as reported clinically in the literature [4,14,24-27].

2. Methods

2.1. Participants

Participants were children referred from the Occupational Therapy (OT) Department at The Children's Hospital in Denver, CO, with a clinical diagnosis of SMD. Children were referred to OT before being recruited for this study. Referrals to OT were made by physicians, teachers, and parents based on aggressive or withdrawn behavior, sensory or motor problems, inattention and impulsivity, and other behaviors disrupting activities of daily living. The identification of SMD was proffered after referral to OT based on global clinical impression after a 2- to 3-hour comprehensive OT evaluation with an advanced clinician. When no criterion standard assessment of a disorder is available, clinicians often rely on their clinical judgment. Global clinical impression of SMD requires the OT to interpret the qualitative information collected during an evaluation within the context of the client's presenting problems to determine whether the presently symptoms are indicative of SMD or another disorder. In this study, global clinical impression was based on interpretation of results from a standardized norm-referenced scale of sensory-motor abilities (ie, the Sensory Integration and Praxis Test), standard clinical observations in an OT gym, and a detailed sensory, developmental, and medical history interview with parents. Symptoms of SMD were also recorded on the "SMD Behavior During Testing Checklist" [28,29], which was completed by the evaluating OT and contributed to the global clinical impression. One hundred forty-three children were identified with significant symptoms of SMD. Forty-four children were excluded due to comorbid diagnoses, for example, cerebral palsy, fetal alcohol syndrome, autistic spectrum disorder, fragile × syndrome, Tourette disorder, or significant cognitive delay. Five parents chose not to have their children enrolled in the study and were excluded. The final sample included 94 children; demographic data by 3 age groups are presented in Table 1.

Recruited subjects were provided with written and verbal information about study procedures, and an informed consent was obtained from all parents (and assent from children older than 7 years) before participation in the study. Consent forms and procedures were approved by the Combined Internal Review Board of the University of Colorado Denver and The Children's Hospital of Denver.

2.2. Instrumentation

One parent of each child completed 4 standardized parent-report measures.

The *Short Sensory Profile (SSP)* [30,31] is a 38-item measure of functional behavior associated with responses to sensory stimuli. Parents indicate their perception of the frequency with which their child exhibits atypical behaviors in response to sensory stimulation using the following: 1 (always) to 5 (never rating). The scale evaluates tactile, visual/auditory, taste/smell, and movement sensitivity; auditory filtering; low energy/weak; and sensation seeking. Raw scores range from 38 to 190 with higher scores reflecting more typical (eg, normal) behaviors. Short Sensory Profile norms were determined by extrapolation from the normative sample of the Sensory Profile [12,13], which was standardized on 1200 children ages 3 to 10 years, stratified by age and sex. Reliability of the SSP subtests range from 0.82 to 0.89 [31].

The *Child Behavior Checklist* (CBCL; form for ages 4-18 years) [32] is a 118-item evaluation of parent perception of children's emotional and behavioral competency. Parents rate how true each item is now or within the past 6 months for their child on a scale from 0 (not true) to 2 (very or often true). High scores indicate more problematic behaviors. Item scores are summarized into 8 clinical subtests, of which all 8 were relevant to the study: withdrawn, somatic, anxious/depression, social problems, thought problems, attention problems, delinquent, and aggressive behavior. These subtests are further summarized into 2 major categories: internalizing problems and externalizing problems. Child Behavior Checklist–scaled scores are based on principal component analyses of parents' ratings of 4455 clinically referred children and normalized on 2368 children ages 4 to 18 years. The normative sample was nationally stratified on socioeconomic status, ethnicity, geographic region, and urban-suburban-rural residence. Test-retest reliability correlation for total scale is 0.93, and interrater reliability is 0.76 [32]. Test-retest reliability coefficients for subtests range from 0.95 to 1.0; internal consistency reliability ranges from 0.78 to 0.97; interrater reliability ranges from 0.93 to 0.96 [32].

The *ADD-H Comprehensive Teacher's Rating Scale* (ACTeRS) [33-35] evaluates attention and hyperactivity based on parent report. Behaviors are rated on a 5-point scale for 24 items, which are collapsed into 4 factors, of which 3 were used in this study: hyperactivity, social skills, and attention. The ACTeRS norms were based on 1399 children. Test-retest reliability coefficients for subtests range from 0.77 to 0.83; internal consistency reliability ranges from 0.91 to 0.98; interrater reliability ranges from 0.50 to 0.61 [34]. On the ACTeRS, high scores indicate normal performance on 2 subtests (hyperactivity and oppositional) and atypical behavior on the other 2 subtests (attention and social skills) [35].

Leiter International Performance Scale–Revised: Parent Rating Scale (Leiter-R PRS) [36] is an instrument that assesses parent's perception of the child's attentional and emotional capabilities—attention, hyperactivity, impulsivity, adaptation, socialization, energy and feelings, moods and confidence, sensitivity/regulation, and socialization—as well as 2 summary subtests: cognitive/social functioning and emotion regulation. Item scores range from 1 to 3, with higher values indicative of more typical (normal) behavior. Standardized subtest scores range from 1 to 19 (mean ± SD, 10 ± 3). The Leiter-R PRS norms were derived from a representative sample of 785 children ages 2 to 21 years, stratified by age, sex, and socioeconomic factors. Subtest reliability ranges from 0.79 to 0.97 [36].

2.3. Data analysis

2.3.1. Equating subtest scores

Each subject had multiple subtest scores from the 4 instruments: 7 from the SSP, 8 from the CBCL, 2 from the ACTeRS, and 8 from the Leiter-R PRS (plus 4 composite scores, 2 each from the CBCL and Leiter-R PRS). Some subjects did not have complete scores for all subtests because of a parent failing to provide an answer for all questions. As part of this study, subtest scores from each instrument were range standardized to have equitable scores across each subtest. Subtest scores were range standardized by subtracting the score from the population maximum and dividing by the difference between the population maximum and minimum resulting in a scale of 0 to 1, with 0 representing an abnormal score and 1 representing a normal score. Range

Table 1
Demographic characteristics of participants

Age groups (y)	Boys	Girls	White	Minority	Total
4-7	33	19	42	10	52
7-10	24	12	34	2	36
10-14	4	2	6	0	6
Total	61	33	82	12	94

standardized scores from the CBCL and ACTeRS were subtracted from 1 because numbers close to 1 on these instruments indicate poor performance, and for this study, it was preferred to have higher values representing better performance. Two standard deviations below the mean was the cut point selected for impaired function.

2.3.2. Subtest aggregation

Subtests in 2 categories, attention and hyperactivity, appeared in more than 1 instrument and measured the same abilities; hence, the scores were collapsed into 1 construct. For example, attention was assessed with the ACTeRS (n = 57), Leiter-R PRS (n = 87), and the CBCL (n = 57). Attention subtests from each instrument were highly correlated ($r = 0.49$; $P < .0001$) and therefore were averaged to create a single attention variable for each subject. Hyperactivity was assessed with the Leiter-R PRS (n = 87) and the ACTeRS (n = 57). Hyperactivity subtest scores from the 2 instruments were significantly correlated ($r = 0.30$; $P < .05$) and therefore were averaged to create a single hyperactivity variable for each subject.

2.3.3. Subtest designation to domains

A qualitative content analysis was completed for each subtest to determine which of the 3 primary domains identified in the literature (attention, sensation, emotion) best represented the constructs (questions) of the subtest. Subtest scores within each domain were highly correlated ($r > 0.70$). Subtest scores across domains were less correlated ($0.31 < r < 0.63$). This validated that the aggregation of subtests within each domain in which they were subjectively placed was correct.

2.3.4. Selection of explanatory variables

Explanatory variables were selected based on distribution characteristics shown to ensure the generalizability of the results to other samples of the population [37]. The 3 specific data characteristics desired for explanatory variables in cluster analysis were examined to determine which variables best fit the criteria for explanatory variables: (1) variables with complete data, (2) variables with data that demonstrated a multimodal distribution, and (3) variables with data that demonstrated positive intervariable correlations. To identify which of the subtests (variables) in this study were the best candidate explanatory variables, we completed several descriptive analyses. First, variables with 10% or more missing data were excluded (n = 9), because records with missing data will be excluded from the statistical analysis. Second, variable distributions with outliers (greater than 2 SD away from the mean) were excluded (n = 5), because data with many outliers are unfavorable for cluster analysis [38]. Third, scatter-plot matrices were examined to identify natural groupings that existed within the distributions of each variable. At this point, 4 variables were identified as possible explanatory variables: hyperactivity, movement sensitivity, auditory filtering, and seeks sensation. Last, the distribution modality was evaluated for multimodal distributions that suggest the presence of cluster structures within the variable. Application of this last descriptive analysis resulted in the identification of 2 variables as the best candidates for explanatory variables: hyperactivity and movement sensitivity.

2.3.5. Clustering decisions

SAS System 8.1 (SAS, Cary, NC) was used to perform Ward's minimum variance hierarchical clustering with Euclidean distances to identify potential clusters in the cohort. Ward's analysis was selected, because it is a nonoverlapping and agglomerative method, which has good cluster recovery ability and has performed well with behavioral data [37,38].

Three internal clustering statistics were used as statistical indices: the cubic clustering criterion, the pseudo-F, and the pseudo-t^2. The agreement of these 3 statistics is suggested as the strongest estimator for the number of cluster structures in the data set [39]. Consensus of the 3 statistics is defined as small pseudo-t^2 values confirmed by peaks in cubic clustering criterion and pseudo-F.

2.3.6. External analysis to discriminate the clusters

The agreement between the cluster solutions and the hypothesized subtypes of SMD was evaluated using subtest scores and demographic variables external to the cluster analysis, which had been originally selected for clinical relevance in evaluating attention, sensation, and emotion: Leiter-R PRS, SSP, ACTeRS, and CBCL. . These included 5 sensation, 4 attention, and 7 emotion subtests (including 2 composite scores). The average for each subtest score was calculated across all individuals within each cluster, as defined by the explanatory variables (hyperactivity and movement sensitivity). The mean scores for each subtest and demographic variables (age, race, sex) were statistically compared using t test. To protect against inflation of type 1 error from multiple t tests, $P < .001$ was used as the critical value for significance. Subtest scores that were significantly different were used to characterize each cluster.

3. Results

3.1. Cluster identification

With hyperactivity and movement sensitivity as the explanatory variables, the strongest agreement between the 3 clustering statistics occurred with 2 clusters: cluster 1 (n = 72) and cluster 2 (n = 22). The former reflected problems with hyperactivity, and the latter reflected problems with movement sensitivity.

Fig. 1 display the hyperactivity and movement sensitivity range standardized mean subtest scores for each cluster. Values closer to 1 represent more typical (normal) performance, and values closer to 0 represent more atypical (abnormal) performance. Thus, cluster 1 has a lower hyperactivity score reflecting atypical performance, whereas cluster 2 demonstrates a higher score reflecting more normal (typical) performance ($t = 3.7$, $P < .0001$).

Table 2
Demographic characteristics for clusters 1 and 2

	Age (y)		Sex		Race	
Cluster	Mean	SD	Male	Female	White	Minority
1 (n = 72)	7.11	2.02	51	21	62	10
2 (n = 22)	6.85	1.83	16	6	21	11

The results demonstrate that cluster 1 (n = 72) has more hyperactive characteristics but less movement sensitivity; cluster 2 (n = 22) has more movement sensitivity with less hyperactivity. We examined the demographic data across the 2 clusters and found no significant difference in mean age or distributions of sex or race (Table 2).

3.2. Subtest aggregation into domains

Each subtest was categorized based on content (Table 3). The sensation subtests were movement, tactile, taste/smell, and visual/auditory sensitivity, and low energy/weak from the SSP and sensitivity and regulation from the Leiter-R PRS. These sensory subtests were highly correlated with each other ($0.70 < r < 0.86$, $P = .04$). The attention subtests were hyperactivity, impulsivity and attention from the Leiter-R PRS and CBCL, and seeks sensation and auditory filtering from the SSP. These subtests were correlated ($0.52 < r < 0.8$, $P = .04$). Finally, the emotion subtests were delinquent, aggressive, socialization, adaptation, and withdrawal from the Leiter-R PRS and the CBCL. These subtests were correlated with each other ($0.49 < r < 0.78$; $P = .04$).

3.3. External analysis-cluster characterization based on the 3 primary domains

Cluster 2 has significantly more movement sensitivity and is significantly more impaired in the sensory subtest lower energy/weak ($P < .0001$) than cluster 1 (Fig. 2). Cluster 1 has significantly more impulsivity ($P < .0001$), inattention ($P < .0001$), and abnormal sensation seeking/craving characteristics ($P < .0001$) than does cluster 2 (Fig. 3). Cluster 1 has more aggressive and delinquent characteristics with more impaired socialization and adaptation skills ($P < .0001$) than does cluster 2. However, cluster 2 is more withdrawn ($P < .0001$) than cluster 1 (Fig. 4). Results from the CBCL and Leiter Emotion composite scores indicate that cluster 1 has more impaired externalizing emotions and cognitive/social impairments ($P < .0001$), whereas cluster 2 demonstrates more impaired internalizing ($P < .05$) (Fig. 5).

Table 3
Grouping of all subtests by the 3 content domains

Sensation subtests	Attention subtests	Emotion subtests
Movement sensitivity	Hyperactivity	Delinquent
Tactile sensitivity	Impulsivity	Aggressive
Audio/visual sensitivity	Seeks sensation	Socialization
Taste/smell sensitivity	Auditory Filtering	Withdrawal
Sensitivity regulation	Attention	Adaptation
Low energy		

4. Discussion

The main finding from this study is that 2 distinct clusters of SMD exist in this sample, potentially representing 2 subtypes of SMD [5]. In this cohort of children with SMD cluster 1, the first subtype includes 75% of the sample (n = 72) and is marked by the explanatory variable hyperactivity. Additional defining characteristics include sensory seeking/craving, impulsivity, delinquent attributes, aggressiveness, poor socialization, poor adaptation, impaired cognitive/social, and

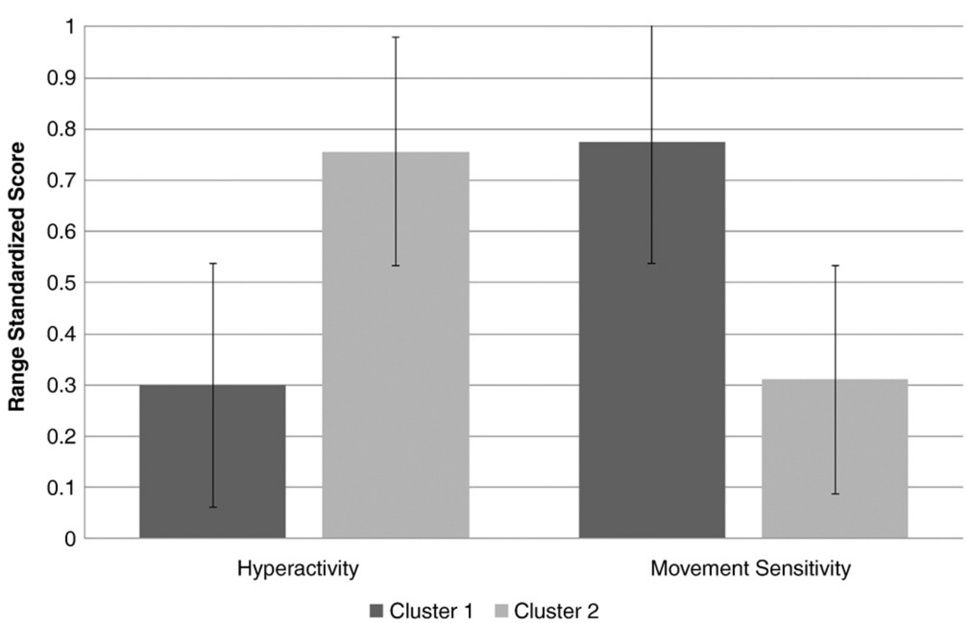

Fig. 1. Range standardized scores for explanatory variables by cluster.

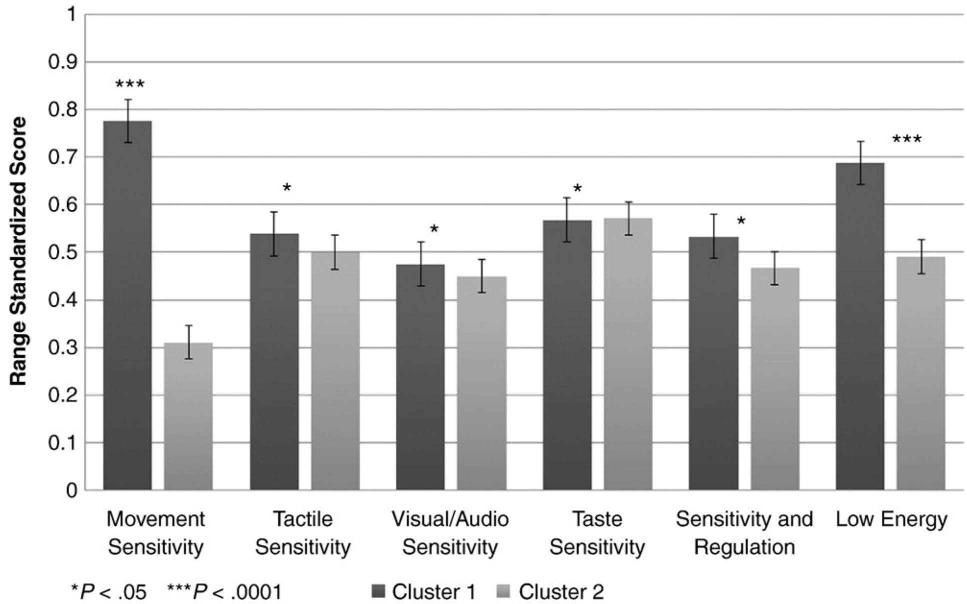

Fig. 2. Sensory domain subtest scores by cluster.

many externalizing behaviors. These characteristics describe the SMD subtype called *sensory seeking/craving*. The item content of the sensory seeking/craving subtest of the SSP overlaps to a great degree with items in the *Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition* describing ADHD particularly on items measuring of attention and impulsivity. Sensory-seeking/craving behavior may be misinterpreted as hyperactivity. Clinically children with sensory seeking/craving calm down when provided with sensory-based activities, such as sitting on a large therapy ball so that they receive proprioceptive and vestibular input. However, children with ADHD do not benefit from this input, but rather get more wound up and disorganized with sensory input. In addition, stimulant medications appear to work well with children classified as ADHD [40-43], but less well with children who have significant sensory-seeking behaviors [44,45]. Impulsivity and hyperactivity behaviors may look similar in children with ADHD compared with children with sensory seeking/craving, but we hypothesize that they are based on different neural mechanisms.

It is important to note that children with ADHD are a heterogeneous group that likely presents with more than 1

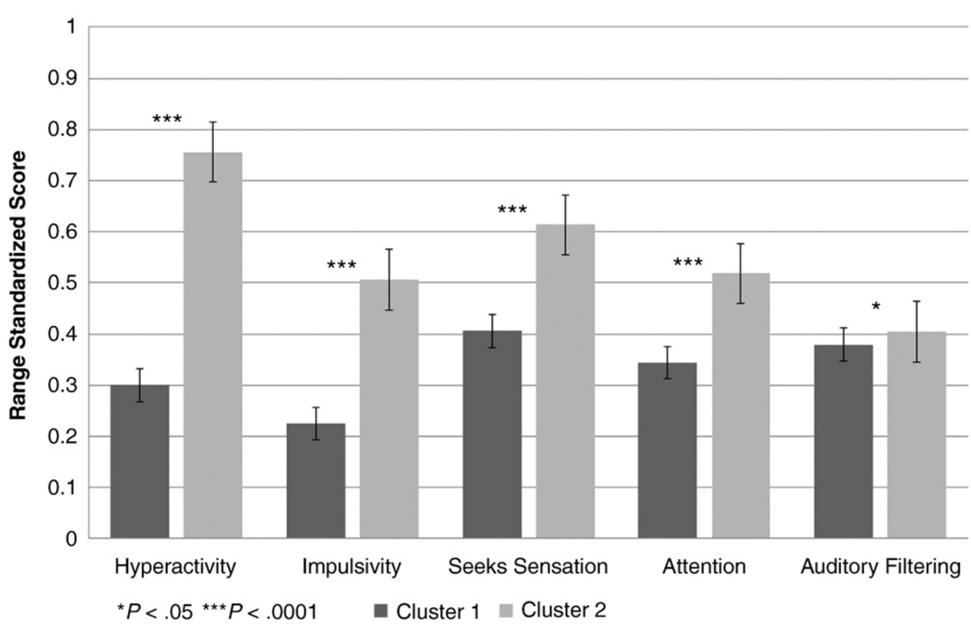

Fig. 3. Attention domain subtest scores by cluster.

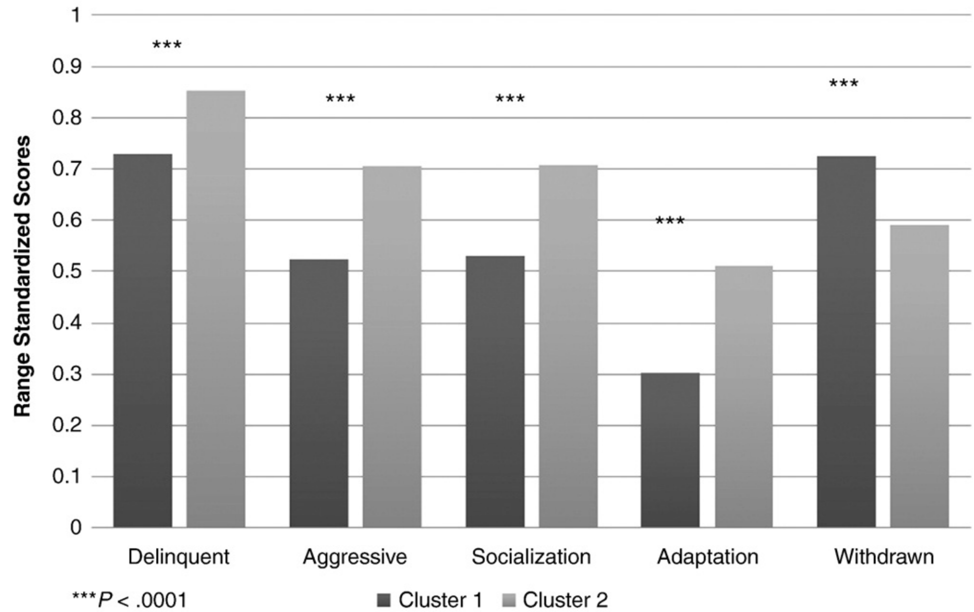

Fig. 4. Emotion domain subtest scores by cluster.

subtype of SMD. In a recent study, Lane and colleagues [46] identified a group of children with ADHD who had behaviors reflective of sensory overresponsivity and had high levels of comorbid anxiety. It is possible that identifying comorbid SMD subtypes will help describe some of the heterogeneity in this diagnostic group. Clearly, children with hyperactivity, impulsivity, and attention problems should be evaluated for SMD as well as ADHD.

Cluster 2, the second subtype, accounts for 25% of the sample (n = 22) and is marked by the explanatory variable movement sensitivity. Additional defining characteristics include withdrawn and low energy/weak behaviors. The low energy/weak behavior associated with this cluster is not surprising given the sensitivity to movement in this subtype. Individuals with low energy/weak behaviors tend to have muscle weakness and muscle fatigue (difficulty sustaining muscle activation). Clinically poor balance and motor control are often associated with low energy/weak behaviors. Thus, sensitivity to movement may co-occur because these individuals frequently avoid activities that challenge their balance system. These characteristics partially describe the subtype of SMD called *sensory underresponsivity*.

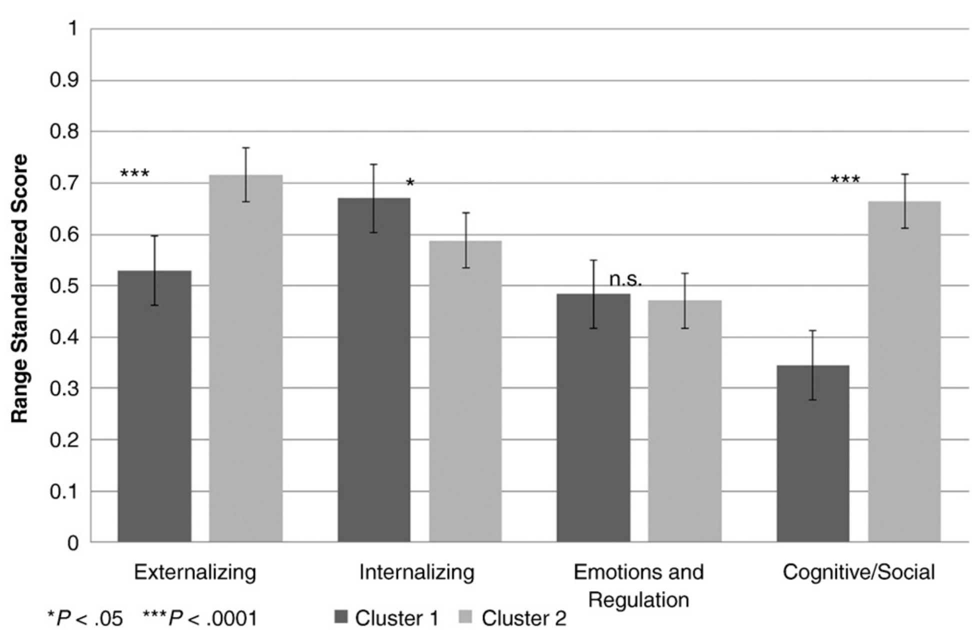

Fig. 5. Emotion composite subtest scores by cluster.

The finding of these particular 2 clusters in the current sample partially supports the new taxonomy proposed by Miller and colleagues [5], which delineates 3 subtypes of SMD: sensory overresponsivity, sensory underresponsivity, and sensory seeking/craving. The cluster analysis differentiated 2 patterns that seem consistent with sensory seeking/craving and sensory underresponsivity. Interestingly, in this sample, behaviors from the third proposed subtype, sensory overresponsive (eg, oversensitivity to touch, visual, auditory, taste, and smell) appeared in both clusters 1 and 2, for example, similar tactile, visual/auditory, and taste sensitivities. Sensory seekers/cravers are described as individuals who have an intense need to enhance their daily sensory experiences. Dunn [12,13,47,48] theorizes that sensory seekers have a high threshold for detecting sensory stimuli (ie, do not notice stimuli easily) and use an active strategy of self-regulation. She compares this with a poor registration group who she hypothesizes also have a high threshold for perceiving sensory information, but use a passive strategy of self-regulation.

The findings in this study are different than the model proposed by Dunn [12,13,47,48]. The sensory seekers in this study did not exhibit problems on the low energy-weak subtest of the SSP, suggesting that they did not have sensory underresponsivity in the proprioceptive and vestibular domains. In addition, the results of this study are different than other previous hypotheses, which suggest that sensory underresponsivity and sensory overresponsivity are on a continuum with [49]. In fact, some individuals in cluster 2 with sensory underresponsivity also had clinically significant sensory overresponsivity (eg, movement sensitivity), suggesting that sensory underresponsivity and movement overresponsivity may co-occur in a group of children. Dopheide [42] and Royeen and Lane [50] suggest that some individuals may manifest both sensory overresponsivity and sensory underresponsivity, within the same sensory domain. Because the SSP only measures underresponsivity on 1 subtest low energy/weak, whether individuals in cluster 2 were underresponsive in other sensory domains remains unclear. Further study is needed to answer this question using tools that use more comprehensive measures of sensory underresponsivity, sensory overresponsivity, and sensory seeking/craving in all 7 sensory domains.

Related to this study finding that 75% of the SMD sample had some sensory seeking/craving and hyperactivity characteristics, it is interesting to note that a high incidence of SMD is reported (82%) in children with ADHD [51]. An empirical prospective study of the sensory symptoms in children with ADHD has yet to be completed; however, as early as 1964, an association between tactile overresponsivity and hyperactive, distractible behaviors was noted in the literature [5]. More recently, a psychophysiologic study of children with ADHD, using somatosensory-evoked potentials, demonstrated that a large percentage of children with ADHD have tactile overresponsivity (a characteristic of the SMD subtype sensory overresponsivity) [52,53]. These findings, in combination with the data from the current study, suggest that a substantial group of children with ADHD have concomitant SMD and vice versa. Clearly, more research is needed to clarify this issue.

The data in this study also suggest that further empirical exploration of the boundaries between ADHD and SMD is important, given overlapping symptoms. Increasingly, researchers are suggesting that disorders should be discriminated with biologic markers. In this regard, preliminary data show that one subtype of SMD (sensory overresponsiveness) shows a characteristic habituation pattern (eg, poor habituation to sensory stimuli) and increased response magnitudes compared with typically developing controls as measured by electrodermal activity during a sensory challenge [31,53]. These data provide some preliminary evidence that there may be a biologic marker for this subtype of SMD. However, further research is needed to validate these data and determine if electrodermal activity is a useful biologic marker of SMD and whether individuals with sensory overresponsivity and sensory seeking/craving have patterns of electrodermal activity that differentiate them from other subtypes and other clinical diagnosis.

Currently, few mental health and developmental disorders are diagnosed from biologic markers. Rather, diagnoses are based on global clinical impressions that follow a specific taxonomy such as the *Diagnostic and Statistical Manual of Mental Disorder*, *Fourth Edition, Text Revision* [54] or the *International Statistical Classification of Diseases and Related Health Problems-9* [55]. However, these descriptive dichotomous classification models fall short in accurately delineating boundaries between disorders and are inelegant in adequately describing the complexity of many clinical disorders [54,55].

5. Limitations

Limitations exist in this study. First, the cohort was patients referred to an OT clinic and may not generalize to children not referred to occupational therapy clinics, including those children with SMD that go undiagnosed. Children referred to other OT clinics could have characteristics that are not the same as this sample; for example,., they may have fewer or more sensory impairments, have more comorbidities, be of different socioeconomic status, or have environmental factors that differentiate them from the sample in this study.

Second, data used in this study are based on parent reporting. Confirmation of these results using direct performance tests or physiologic measures of sensory responsivity would be useful to cross-validate these findings.

6. Conclusions

The findings of the 2 clusters of SMD in the sample studied suggest that SMD subtypes include at least 2 distinct subtypes: sensory seeking/craving as determined by cluster 1 (subtype 1) and sensory underresponsivity as determined by cluster 2 (subtype 2). This is the first empirical study to differentiate sensory seeking and sensory underresponsivity, which are typically thought to be the same clinically, due in part to the Dunn model that identifies these 2 subtypes as falling on the same behavioral spectrum, with one being passive and the other being active. Further differentiating sensory modulation subtypes can aid researchers in designing studies with homogeneous samples and can aid therapists in designing effective intervention protocols for children with sensory challenges. Clustering methods are useful to view subtype characteristics especially, as in this case, when the true grouping is unknown and is based on clinical impressions only. This study moves the field a step forward in understanding the complexities of children with SMDs. Further research is needed to cross-validate results presented here and to further define the components of SMD. Future research should investigate methods to assess sensory underresponsive characteristics in children because current parent-report measures focus primarily on sensory over-responsivity and sensory-seeking/craving attributes.

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