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Is there a relationship between restricted, repetitive, stereotyped behaviors and interests and abnormal sensory response in children with autism spectrum disorders?

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Abstract

This study examined the relation between restricted, repetitive, and stereotyped behaviors and interests (RBs) and sensory responses in a group of 70 children and adolescents diagnosed with an autism spectrum disorder (ASD). Caregivers completed the Repetitive Behavior Scale-Revised (RBS-R) and the Sensory Profile. Controlling for IQ and age, total RBS-R and Sensory Profile scores revealed significant correlations both prior to and after removing overlapping items. Examination of the co-occurrence of RBs and atypical sensory responses in this population suggests a subgroup has consistently high rates of problems in both RBs and sensory processing. In addition, this subgroup has high rates of prescribed psychoactive medications and co-morbid psychiatric diagnoses. The IQ and age of this subgroup did not differ significantly from the rest of the participants. Results are consistent with previous research describing the co-occurrence of RBs and sensory response abnormalities in the ASD population. Further investigation of the subset of individuals with ASD who have high rates of RBs and abnormal sensory responses may lead to a more comprehensive

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understanding of their clinical picture and improve interventions. Additionally, research with this subgroup may have significance for identifying a specific phenotype in ASD.

Keywords: Repetitive behaviors; Sensory response; Autism spectrum disorders; Children; Adolescents

1. Introduction

Autism spectrum disorders (ASD) include Autistic Disorder, Asperger's Disorder, and Pervasive Developmental Disorder, Not Otherwise Specified (American Psychiatric Association, 1994). The term ''spectrum disorders'' is commonly used to refer to this population and has been defined as ''. . . a group of disorders that are thought to be related through the sharing of risk genes or pathophysiological mechanisms'' (Hyman, 2007, p. 730). ASDs are diagnosed based on a core triad of clinically observable symptoms involving impaired social interaction and communication abilities along with restricted, repetitive, and stereotyped behaviors and interests (RBs) (American Psychiatric Association, 1994). Specifically, RBs include stereotyped and repetitive body movements and manipulation of object parts; compulsive or ritualized behaviors; insistence on sameness of the environment and routines; circumscribed interests, and self-injurious behaviors (Bodfish, Symons, Parker, & Lewis, 2000; Lewis & Bodfish, 1998; Schultz & Berkson, 1995). RB features in individuals with ASD vary in their occurrence, frequency, and severity (Bodfish et al., 2000). RB features also appear as a part of early typical development, are present in individuals with other developmental disabilities (Berkson, 2002; Berkson, Tupa, & Sherman, 2001), and are a diagnostic part of other mental disorders, such as Obsessive Compulsive Disorder (American Psychiatric Association, 1994). Some reports suggest that intellectual ability is highly correlated with both the occurrence and type of RBs in individuals with autism (Bartak & Rutter, 1976; Carcani-Rathwell, Rabe-Hasketh, & Santosh, 2006; Gabriels, Cuccaro, Hill, Ivers, & Goldson, 2005; Matson, Kiely, & Bamburg, 1997; Militerni, Bravaccio, Falco, Fico, & Palermo, 2002; Poustka & Lisch, 1993; Thompson & Berkson, 1985); however, if a specific pattern of RBs distinguishes a unique ASD phenotype from general cognitive disabilities is not known.

In addition to the core diagnostic features, individuals with ASD have a variety of challenges in areas such as cognition (Chakrabarti & Fombonne, 2001; Lainhart, 2003; Yeargin-Allsopp et al., 2003), adaptive behavior (Gabriels, Ivers, Hill, Agnew, & McNeill, 2007), and sleep (Schreck, Mulick, & Smith, 2004). They also may have seizures (Elia, Musumeci, Ferri, & Bergonzi, 1995; Tuchman, Rapin, & Shinnar, 1991) and/or co-morbid psychiatric disorders (Bradley, Summers, Wood, & Bryson, 2004; Leyfer et al., 2006; Sverd, 2003). Numerous studies report clinical evidence of abnormal responses to sensory stimuli in the ASD population (Baranek, Foster, & Berkson, 1997; Gillberg et al., 1990; Kern et al., 2006; Kern et al., 2007; Liss, Saulnier, Fein, & Kinsbourne, 2006; Ornitz, Guthrie, & Farley, 1977; Ornitz, Guthrie, & Farley, 1978; Tecchio et al., 2003; Tomchek & Dunn, 2007; Volkmar, Cohen, & Paul, 1986; Wainwright-Sharp & Bryson, 1993) including over-responsivity, under-responsivity and sensory seeking behavior. All seven sensory domains may be affected particularly auditory, visual, vestibular, tactile, and proprioceptive domains (Dunn, 1999). Atypical sensory response patterns occur in many individuals with autism (Baranek, David, Poe, Stone, & Watson, 2005; Greenspan & Wieder, 1997; Hirstein, Iversen, & Ramachandran, 2001; Tomchek & Dunn, 2007) and together all three subtypes are known as Sensory Modulation Disorder, which is associated with abnormal

arousal. Studies using parent-report scales suggest that children with autism have significantly more abnormal sensory responses than do children with other developmental delays and/or typically developing children (Baranek, Parham, & Bodfish, 2005; Dahlgren & Gillberg, 1989; Kientz & Dunn, 1997; Lord, 1995; Lord, Rutter, & Le Couteur, 1994; Ornitz et al., 1978; Rogers, Hepburn, & Wehner, 2003; Watling, Deitz, & White, 2001). Similarly high levels of sensory responsiveness are noted in individuals with fragile X syndrome, receptive aphasia, and deaf-blindness (Miller, Polatajko, Missiuna, Mandich, & Macnab, 2001; Rogers et al., 2003; Wing, 1969). However, a limitation in many previous studies is information specifying the cognitive level of the sample and what the relation is between cognition and sensory response.

Researchers suggests two disparate reasons that individuals with ASD might engage in RBs: (1) to induce a sensory experience and (2) as reaction to sensory stimulation (Liss et al., 2006). Studies have examined specifically this relationship between RBs and sensory response to environmental stimulation in individuals with developmental disabilities and those with autism (Baranek et al., 1997; Colman, Frankel, Ritvo, & Freeman, 1976; Gal, Dyck, & Passmore, 2002; Grandin, 1992; Willemsen-Swinkels, Buitelaar, Dekker, & van Engeland, 1998). For example, tactile over-responsivity (i.e., aversive responses to tactile stimulation which is not noxious to most people) has been associated with more rigid stereotyped behaviors (e.g. insistence on sameness and repetitive verbalizations) (Baranek et al., 1997). Visual over-responsivity and increased RBs were noted with florescent light compared to incandescent lighting in ASD (Colman et al., 1976). Further studies have shown that ''attractive'' sensory stimuli are related to reduced stereotyped movements compared to ''aversive'' sensory stimuli (Gal et al., 2002) in children with autism and MR (IQs < 50). Despite these findings that suggest a direct relation between RBs and abnormal sensory responsiveness in individuals, research on measurement tools which use both RBs and sensory items is lacking rigor. Thus certain scales label a behavior as an RB while different scales label the same behavior as sensory. The inconsistent labeling of behavior confounds interpretation of the overlap/discrimination of RBs and sensory response abnormalities. For example, behavior labeled repetitive movement behaviors in the DSM-IV (American Psychiatric Association, 1994) are used interchangeably with ''sensory seeking'' behaviors (Liss et al., 2006). One scale, the Short Sensory Profile (McIntosh, Miller, Shyu, & Hagerman, 1999) classifies ''touches people and objects'' as a sensory seeking behavior, and yet another scale, the Repetitive Behavior Scale-Revised (Bodfish et al., 1999) classifies the same behavior as a compulsive behavior.

The aim of this pilot study was to evaluate whether a relationship existed between RBs and sensory response in children with ASD. Secondarily, the goal was elucidating whether the association is due to item overlap in scales. The third goal was examining whether a phenotypic subtype of ASD was suggested with high rates of RBs and abnormal sensory responses. For all research questions methodology used in previous studies was improved upon by measuring and controlling for IQ, since intellectual ability has been shown to be related to the expression of RBs (Bartak & Rutter, 1976; Carcani-Rathwell et al., 2006; Gabriels et al., 2005; Matson et al., 1997; Militerni et al., 2002; Poustka & Lisch, 1993; Thompson & Berkson, 1985).

2. Methods

2.1. Participants

Children with a clinical diagnosis of an ASD ($n = 70$; 58 males, 12 females) participated. The sample was recruited from clinical, research, and community settings. The mean age of all

participants in this study was 10.8 ± 4. 0 years (range: 3.0–19.7 years) and the average IQ was 81.4 ± 26.1 (range 25–138). Forty-two of the 70 participants (60%) were taking psychotropic medications at the time of the study, including atypical antipsychotics, mood stabilizers, SSRIs, stimulants, opioid agonists and alpha adrenergic agonists. Thirty (43%) of the 70 participants were diagnosed with co-morbid psychiatric disorders including mood disorders, anxiety disorders, attention deficit disorders, psychotic disorders, and sleep disorders. Thirty-one (44%) of the 70 participants were identified as pubescent by their caregivers.

Inclusion criteria were: (a) documentation of participants' full scale IQ (within 41 months of study entry); and (b) diagnostic data to confirm a DSM-IV (American Psychiatric Association, 1994) clinical diagnosis of an ASD. IQ tests review included the Wechsler Intelligence Scales (Wechsler, 1981, 1989, 1991, 1997, 2003), the Bayley Scales of Infant Development (Bayley, 1993), the Mullen Scales of Early Learning (Mullen, 1995), the Leiter International Performance Scale-R (Roid & Miller, 1997), the Kaufman-ABC (Kaufman & Kaufman, 1983), and the Differential Abilities Scales (Elliott, Murray, & Pearson, 1990) (average full scale IQ = 81.4, ranging from 25 to 138). In addition, caregivers responded to a screening, which identified the puberty status of their child based on Tanner's criteria (Tanner, 1962) (e.g., physically observable pubertal maturation features such as genital and pubic hair in boys and breast and pubic hair in girls. In this sample, the five Tanner stages were divided into two groupings: Group 1 (pre-pubescent) and Group 2: all other Tanner stages 2–5 (pubescent). Subject demographics are summarized in Table 1.

2.2. Instrumentation

2.2.1. Repetitive Behavior Scale-Revised (RBS-R)

The RBS-R (Bodfish et al., 1999) is an empirically derived 43-item caregiver report of the full spectrum of RBs, consisting of six distinct subscales (Stereotyped Behavior, Self-injurious Behavior, Compulsive Behavior, Routine Behavior, Sameness Behavior and Restricted Behavior). The RBS-R is a brief (<15 min to complete), yet comprehensive survey of the entire spectrum of RBs clinically observed and referred to in the DSM-IV (American Psychiatric Association, 1994) diagnostic description of Autistic Disorder. Parents or caregivers rate 43 behaviors on a scale of 0–3, where 0 indicates the behavior does not occur and 3 indicates the behavior does occur and is a severe problem. It showed discriminant validity in adults,

Table 1
Demographic characteristics of participants

	N = 70
Mean age (years)	10.8 (min: 3.0; max: 19.7)
Gender	Male: 58; female: 12
Ethnicity	82.9% Caucasian
Co-morbid psychiatric diagnoses	Yes: 30; no: 40
Puberty	Yes: 31; no: 39
Psychoactive medications	Yes: 42; no: 28
Mean IQ	81.4 (min: 25; max: 138)
Diagnosis	
Autistic Disorder	48
Asperger's Disorder	14
PDD-NOS	8

distinguishing participants with autism and MR from non-autistic participants with MR in the overall RBS severity score (Bodfish et al., 2000).

2.2.2. Sensory Profile (Caregiver Questionnaire)

The Sensory Profile (Dunn, 1999) is a 125-item, 30-min, standard caregiver questionnaire of the effect of sensory processing on the child's ability to function in daily life. Item responses occur on a five point Likert-rating scale from 1 (always occurs) to 5 (never occurs). Normative data for the Sensory Profile were obtained from 1037 typically developing children ages 3–10 years. The developers of this measure have collected data with children with ASD ages 3–17 years (Dunn, 1999). The Sensory Profile provides two sets of standard scores depending on how the items are clustered: (1) domain scores (Sensory Processing, Sensory Modulation, Behavior and Emotional Response) and (2) factor scores (nine empirically derived factors). This study used the Sensory Processing score, a sum of six domain scores (i.e., Auditory, Visual, Vestibular, Touch, Multisensory and Oral Sensory Processing.

2.2.3. Procedure

The study was approved by the Institutional Review Board and caregivers participated in informed consent to review participants' medical records to obtain demographic, diagnostic, and IQ information. Participants were excluded if no standardized IQ results were available. One caregiver for each participant completed both the RBS-R and the Sensory Profile based on the participant's behavior in the past month. Participants' current age, puberty status, and medications were provided by caregivers.

2.2.4. Analytical procedure

Statistical Package for the Social Sciences version 11.5 for Windows (SPSS, 2003) was used for data analysis. For the analyses, alpha was set at 0.05 and a Bonferroni correction for multiple significance tests for the correlation analysis was applied. This reduced the critical p value to $p < 0.007$ to achieve an uncorrected alpha of $p < 0.05$.

To address the primary aim of characterizing the relation between RBs and abnormal sensory response, a partial correlation between RBS-R Total Score and the Sensory Processing domain score was calculated, controlling for age and IQ. To determine whether frequent/severe RBs or sensory processing abnormalities were affected by the use of psychotropic medication, a multivariate analysis of variance (MANOVA) was performed, with dependent factors of total RBS-R score and total Sensory Processing domain score.

3. Results

A significant correlation was found between the RBS-R Total Score and the Sensory Processing domain score (Pearson's $r = -0.61$, $p < 0.001$) with the complete sample. Several items on the RBS-R overlap with items on the Sensory Processing domain of the Sensory Profile. To assure that the relation between the scales was not affected by item overlap, the redundant items from the RBS-R were deleted from the analysis including the complete Stereotyped Behavior Subscale of the RBS-R. Three additional items in the remaining five RBS-R subscales were also dropped. See Table 2 for a list of these specific items. The correlation between the overlapping items from the RBS-R and the Sensory Processing domain of the Sensory Profile was -0.65 ($p < 0.001$), confirming a strong relation among the items.

Table 2
Overlapping measurement items

RBS-R item	Sensory Profile Item
1. Body rocking, body swaying	27. Rocks unconsciously
2. Rolls head, nods head, turns head	24/25. Seeks all kinds of movement (fidgets)
3. Flaps hand, claps hands, shakes hand or arm	24/25. Seeks all kinds of movement (fidgets)
4. Turns in circles, whirls, jumps	26. Twirls/spins self frequently
5. Spins or twirls objects, twiddles or slaps or throws objects, lets objects fall	41. Unusual need to touch certain toys, surfaces or textures
6. Covers eyes, covers ears, smells or sniffs items	15. Covers eyes or squints; 2. Holds hands over ears
12. Rubs or scratches self	39. Rubs or scratches out a spot that have been touched
22. Need to touch, tap or rub items, surfaces or people	40/41/45. Touches people and objects
23. Strongly prefers eating or drinking certain things	61. Shows strong preference for certain tastes

A new analysis with overlapping RBS-R items deleted identified a significant correlation between this shortened RBS-R Total Score and the Sensory Processing domain score of the Sensory Profile (Pearson's $r = -0.53$, $p < 0.001$) with the complete sample. This finding remained relatively unchanged when age and IQ were controlled by partial correlations ($r = -0.53$, $p < 0.001$).

To determine the impact of taking psychoactive medications on the shortened RBS-R Total Score and Sensory Processing domain score of the Sensory Profile, a MANOVA was conducted with full scale IQ as a covariate. The effect of psychoactive medication was non-significant ($p = 0.09$) and inclusion of full scale IQ did not affect the model ($p = 0.07$).

The data were then examined to explore the presence of a phenotypic subgroup within ASD who has both high RBs and atypical sensory responses. The sample was divided into quartiles based on the RBS-R Total Score (with overlapping items deleted) (see Fig. 1) and the upper quartile (''High RB'' subgroup; RBS-R Total Score $\geq$44) compared to the lower three quartiles (''Low RB'' subgroup) using unpaired t-tests and χ^2. No significant differences between the High RB and Low RB subgroups on age ($t = 0.75$, $p = 0.46$), on Full Scale IQ score ($t = 0.21$, $p = 0.84$),

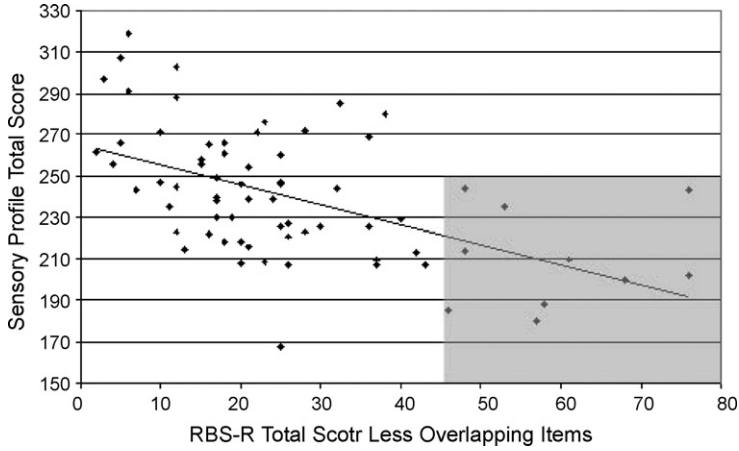

Fig. 1. Sensory Profile Total Score and RBS-R Total Score. (Grey shading in lower right quadrant highlights High RB group.)

Table 3
Characteristics of High RB and Low RB subgroups

	Low RB subgroup ($n = 53$)	High RB subgroup ($n = 17$)	Statistical value	p value
Mean age (years)	10.96 (min: 2.99; max: 17.67)	10.13 (min: 3.63; max: 19.73)	$t = 0.75$	0.46
Mean IQ	81.81 (min: 25; max: 138)	80.29 (min: 38; max: 110)	$t = 0.21$	0.84
RBS-R Total Score	24.02 (min: 2; max: 43)	62.24 (min: 44; max: 95)	$t = 11.29$	<0.001
Sensory Profile Total Score	249.13 (min: 207; max: 319)	212.24 (min: 168; max: 244)	$t = 4.92$	<0.001
Puberty	49.1%	29.4%	$\chi^2 = 2.01$	0.16
Psychoactive medications	52.8%	82.4%	$\chi^2 = 4.67$	0.03
Co-morbid psychiatric diagnoses	35.8%	64.7%	$\chi^2 = 4.38$	0.04

or on puberty status ($\chi^2 = 2.01$, $p = 0.16$) was found. However, the High RB and Low RB subgroups showed significant differences on the Sensory Processing domain of the Sensory Profile Sensory ($t = 4.92$, $p < 0.001$) with the High RB subgroup demonstrating significantly more abnormal sensory responses. Using a χ^2 test, it was shown that the High RB subgroup did not differ significantly from the Low RB group in terms of puberty status ($\chi^2 = 2.01$, $p = 0.16$), but that the High RB subgroup did have a significantly higher percentage of participants taking psychoactive medications ($\chi^2 = 4.67$, $p = 0.03$), 82% in the High RB group compared to 53% in the Low RB subgroup. Co-morbid psychiatric diagnosis were also significantly higher in the High RB subgroup ($\chi^2 = 4.38$, $p = 0.04$) with 64% of the High RB group compared to only 36% of the Low RB group demonstrating additional diagnoses. Examination of the co-morbid diagnoses in the High RB group revealed 59% has a mood disorder, more than half of whom had Bipolar Disorder.

A chart review of the sensory response problems in the High RB subgroup revealed a variety of issues including over, under, and sensory seeking behaviors in relation to visual, tactile, auditory, oral, and vestibular stimulation. Finally, 52% of the participants in the High RB subgroup were not diagnosed with an ASD until at least age six and some were not diagnosed until 18 years of age. See Table 3 for a description of these two subgroups.

4. Discussion

This study demonstrated a significant relationship between frequent/severe RBs, as assessed by the RBS-R Total Score and abnormal sensory responses, as assessed by the Sensory Processing domain of the Sensory Profile, in a group of 70 participants with ASD, controlling for age and IQ. These findings concur with findings from previous smaller studies (Baranek et al., 1997; Colman et al., 1976; Gal et al., 2002; Grandin, 1992; Willemsen-Swinkels et al., 1998). After removing overlapping items from the RBS-R, a significant association remained, which suggests that the relation is not an artifact of item overlap in the dependent measures.

Of particular interest was the suggestion that a subgroup might exist within ASD with high rates of RBs and abnormal sensory responses regardless of age and IQ. The demographic data related to this subgroup suggests that they are more difficult to understand, diagnose, and treat, reflected by more psychoactive medications, diagnosis after age 6 years and more co-morbid psychiatric diagnoses, particularly Bipolar Disorder and other mood disorders. Given the correlation between the RBS-R and the Sensory Processing domain score of the Sensory Profile, it seems logical that the Sensory Processing domain total scores are atypical in the High RB

subgroup. However, the scatter plot in Fig. 1 suggests that participants in the Low RB subgroup have more heterogeneous Sensory Processing domain scores than persons in the High RB subgroup. Specifically, some participants in the Low RB subgroup have minimal abnormal sensory responses. Though this observation is limited by a relatively small sample size, a subgroup with ASD who have high RBs and high atypical sensory responses appears to exist. For example, individuals with autism may be under responsive to pain sensation and cause self-injury by insisting on engaging in repetitive behaviors such as head banging or biting themselves (Gal et al., 2007). This subgroup may benefit from more comprehensive assessment to identify the presence of additional co-morbidities and/or sensory response abnormalities, thus informing clinical interventions leading to a more comprehensive understanding of the clinical picture.

4.1. Limitations of the present study

The findings of this pilot study are limited by the fact that the RB and sensory response measurements were based only on caregiver report measures. Collecting RB and sensory response information from several different reporters across environments would provide a more comprehensive picture of these features. Future studies should also collect reports of RBs and sensory response abnormalities from more than one caregiver including teachers, and if possible, include standardized clinical observations of RBs and abnormal sensory response. This, combined with a larger sample size, would permit more definitive conclusions.

4.2. Future directions

This study suggests more frequent/severe RBs and atypical sensory responses are associated. However, causality cannot be inferred from these correlation findings. Replication studies are needed to support or refute the findings with a larger ASD sample controlling for age, IQ, puberty status, and medication use, as was done in this study. Finally, further research examining whether these two characteristics within autism constitute a consistent phenotypic subgroup is indicated. This research may be significant for better understanding the genetic etiology of ASD.

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