J Autism Dev Disord (2014) 44:3264–3270
DOI 10.1007/s10803-014-2175-x

BRIEF REPORT

Brief Report: Assessment of Early Sensory Processing in Infants at High-Risk of Autism Spectrum Disorder

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Published online: 27 June 2014

Abstract This study assessed sensory processing differences between 24-month infants at high-risk of autism spectrum disorder (ASD), each with an older sibling with ASD, and low-risk infants with no family history of ASD. Sensory processing differences were assessed using the Infant/Toddler Sensory Profile, a parent-reported measure. Groups were compared based on 3-year outcomes: (a) high-risk infants subsequently diagnosed with ASD; (b) high-risk infants without an ASD diagnosis; and (c) low-risk infants without an ASD diagnosis. Analyses showed that high-risk infants diagnosed with ASD have more difficulty with auditory processing (i.e., responses to auditory stimuli) and lower registration (i.e., lacking sensation awareness) compared to controls. Thus, behavioral responses to sensory input represent early risk markers of ASD, particularly in high-risk infants.

Keywords Autism spectrum disorder · Sensory · Infant siblings

Introduction

Sensory processing is the ability to receive, organize, and interpret sensory stimuli, including, but not limited to, oral, visual, tactile, vestibular, and auditory experiences (Dunn 2002). Difficulties with sensory processing (specifically, behavioral responses to sensory input, generally measured by parent report) have been widely reported in children with autism spectrum disorder (ASD) (Ben-Sasson et al. 2007, 2009; O'Donnell et al. 2012). Based on parent- and self-reports, Kern et al. (2006) found abnormalities in auditory, visual, tactile, and oral sensory processing in individuals with ASD aged 3–56 years compared to typically developing controls. Differences appeared to lessen

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with age, although this may have been due to the lack of sensitivity of the measure used to assess sensory processing in adults. Indeed, by incorporating a self-report measure designed for adolescents and adults, Crane et al. (2009) found that sensory processing impairments were highly prevalent in adults with ASD, in comparison to controls, suggesting that sensory issues are relevant to clinical management of ASD across the lifespan.

Based on a systematic review of the literature, Rogers and Ozonoff (2005) reported that sensory symptoms are more frequent in children with ASD, in comparison to typically developing children, but *not* in comparison to other groups of children with disabilities, such as those with Fragile X Syndrome or severe hearing and/or visual impairments. They concluded that sensory symptoms alone cannot be used to differentiate ASD from other disabilities. However, there remains interest in whether behaviors indexing atypical sensory processing may be informative for early detection of ASD. Ben-Sasson et al. (2007) reported that sensory modulation and regulation behaviors were significantly different in toddlers with and without ASD who were matched for mental age. Toddlers with ASD were more likely to be *under-responsive* (i.e., unaware of or slow to respond to sensory input), to display *avoidance* (i.e., limiting the amount and type of sensation), and had a low frequency of *sensory-seeking* behaviors (i.e., seeks out highly stimulating sensory experiences). A case report by Dawson et al. (2000) was among the first to describe the development of a child with ASD who was followed prospectively from early infancy. Particularly prominent were poor state regulation and sensory processing challenges, such as easy startling and hypersensitivity to tactile input in the first 12 months of life.

The Infant/Toddler Sensory Profile (ITSP; Dunn 2002), based on Dunn's Model of Sensory Processing (1997), provides a measurement framework to systematically evaluate toddlers' behavioral responses to sensory stimuli, as observed and reported by parents. The ITSP is comprised of several domains (i.e. subscales), such as auditory processing, visual processing, etc., and four quadrants of sensory responses that are independent from sensory domains (low registration, sensory sensitivity, sensory seeking and sensory avoiding; described below under *Measures*). Using the ITSP, Mulligan and White (2012) compared 13 high-risk (HR) infants (younger siblings of children with ASD), aged 11–13 months, to normative ITSP data (age range between 7 and 36 months), and reported that HR infant siblings had fewer sensory-seeking behaviors, particularly in the auditory processing modality. However, the authors did not examine whether specific sensory processing profiles were associated with ASD outcomes within the HR group. Thus, it is unclear whether differences were related to ASD, broader manifestations of familial risk, or even reporting bias, since parents are not blind to risk status. By stratifying HR sibling samples by diagnostic status at age 3 years, we can clarify whether differences in sensory processing reported early in development are specifically related to ASD. To date, no other prospective reports have assessed sensory processing in HR infants using the ITSP.

The current study aimed to compare 24-month ITSP scores among three groups defined based on diagnostic outcomes at 3 years: (a) HR infants subsequently diagnosed with ASD (HR-ASD); (b) HR infants not diagnosed with ASD (HR-N); and (c) LR infants with no family history of ASD (i.e. no first-degree relative with ASD; LR). It was hypothesized that ITSP scores within each domain would differ significantly among the three groups; in particular, the parents of HR-ASD infants would report more sensory differences in each domain than the parents of the HR-N or LR infants, with HR-N on a gradient between HR-ASD and LR groups.

Methods

Data were collected as part of a multi-site prospective longitudinal study of the early development of HR infants and LR comparison infants (see Zwaigenbaum et al. 2005, 2012 for details). Research ethics approval was received at each of three sites (Hamilton, Halifax, and Toronto). The ITSP was administered at 24 months to assess infants' behavioral responses to a range of sensory inputs, as reported by parents. Diagnostic assessment at 3 years included the Autism Diagnostic Observation Scale (Lord et al. 2002), the Autism Diagnostic Interview – Revised (Lord et al. 1994), and best-estimate clinical diagnosis, informed by these measures. The individual who completed the 36 month diagnostic appointment was blind to previous study assessments. In total, 91 participants contributed complete data; participant demographic information is summarized in Table 1.

Measures

Assessment of Sensory Processing

The ITSP (Dunn 2002) is a 48-item parent-report questionnaire of potential sensory processing difficulties from birth to 36 months of age. Parents rate the frequency of the items from "almost always" (scored as 1) to "almost never" (scored as 5). The ITSP scores assess sensory processing across five domains, including auditory, visual, tactile, vestibular, and oral sensory processing. The ITSP also assesses the child's reaction to the sensory experience

Table 1 Sample demographics and descriptive statistics at 36 months of age

Clinical characteristics	HR-ASD (n = 14)	HR-N (n = 45)	LR (n = 31)
Sex (M:F)	10:4	21:24	14:17
	Mean (SD) Min–Max	Mean (SD) Min–Max	Mean (SD) Min–Max
MSEL—early learning composite	79.15 (20.33)*^	108.68 (20.56)	118.00 (17.15)
Standard score	49–114	61–137	84–149
MSEL—expressive language	39.21 (14.36)*^	52.68 (9.82)	57.77 (8.33)
T-score	19–58	30–70	38–72
MSEL- receptive language	36.71 (12.28)*^	52.23 (9.10)	57.26 (10.88)
T-score	19–53	35–75	35–73
ADOS severity	7.64 (2.02)*^	2.29 (1.62)	2.45 (1.98)

HR-ASD high risk – autism spectrum disorder, *HR-N* high risk – not diagnosed autism spectrum disorder, *n* sample size, *M* male, *F* female, *SD* standard deviation, *MSEL* mullen scales of early learning, *ADOS* autism diagnostic observation schedule

* Different from HR-N ($p < 0.001$); ^ different from LR ($p < 0.001$)

within four quadrants (see Fig. 1): low registration, sensory seeking, sensory sensitivity, and sensation avoiding. If indicated based on initial raw scores, a fifth subscale (or "quadrant"), "low threshold", would be calculated. Domain and quadrant scores are plotted along a continuum, with the "Typical Performance" range comprising ± 1 standard deviation (SD) from the mean of children without disabilities, the "Probable Differences" range comprising scores that fall within 1–2 SDs, and the "Definite Differences" range falls outside 2 SDs of the mean score. Scores are considered clinically significant if they are >1 SD from the mean. The ITSP does not provide an overall score; as such group comparisons in this study were based on domain and quadrant scores. In the normative sample, the reliability of the domain and quadrant scores ranged from 0.69 to 0.85 (Dunn 2002), and good content and criterion validity were reported (Dunn and Daniels 2002).

Autism Symptoms

At 3 years of age, ASD symptoms were measured using two gold-standard tools by a psychometrist, psychologist or developmental pediatrician, trained to research reliability,

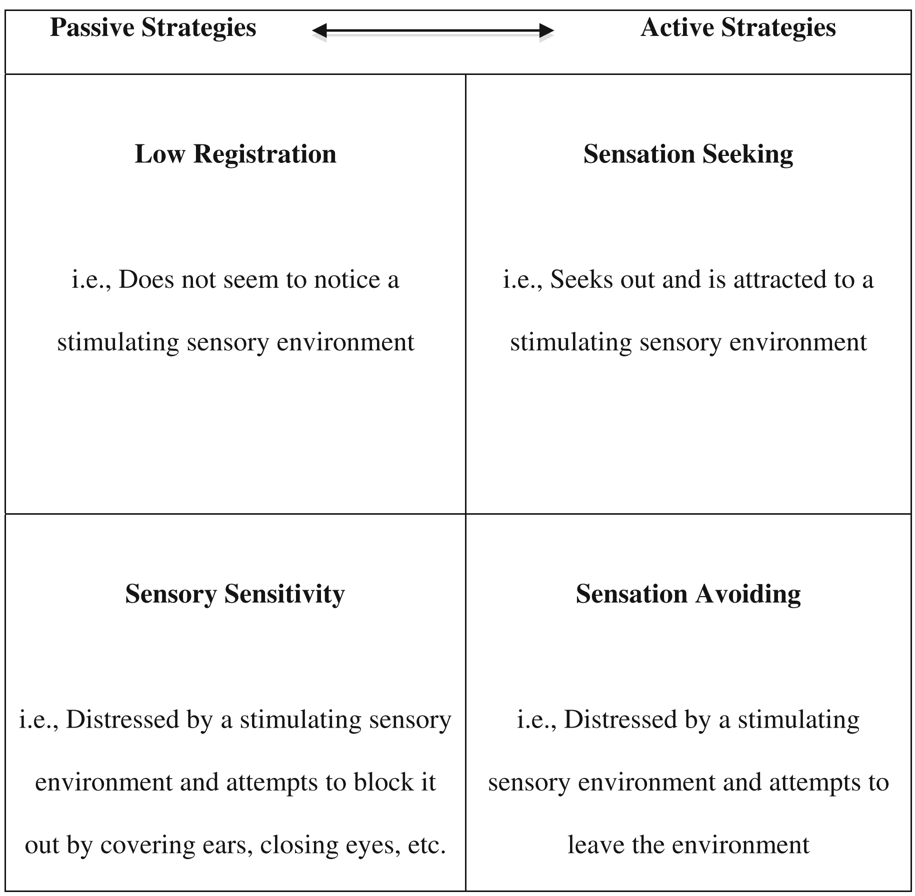

Fig. 1 Self-Regulation Behavioral Response Continuum, modified from Dunn's Model of Sensory Processing (2002). An infant receives a score in each of the four quadrants rated as being: much less than most people, less than most people, similar to most people, more than most people or much more than most people in comparison to other typically developing infants of the same age. This reflects that the reactions to sensory experiences occur across a continuum regardless of sensory domains (i.e., auditory, tactile, vestibular, etc.)

and blind to assessments from previous study visits. The Autism Diagnostic Observation Schedule (ADOS), a semi-structured assessment of communication, social skills, imaginative play, and repetitive behavior (Lord et al. 2002), provided a measure of ASD symptom severity (Gotham et al. 2009). The Autism Diagnostic Interview-Revised (ADI-R) is a semi-structured diagnostic interview that focuses on communication and social development, as well as the presence of repetitive and restricted behaviors (Lord et al. 1994). Some children with a clinical diagnosis of ASD had sub-threshold algorithm scores on the ADOS and/or ADI-R, but met DSM-IV-TR criteria based on expert review of all available 36-month data.

Cognitive Assessment

The Mullen Scales of Early Learning (MSEL; Mullen 1995), which is standardized and validated from birth to 68 months, was used to assess developmental functioning in motor, language, and visual problem-solving domains at each study visit.

Analytic Approach

Groups (LR, HR-N, HR-ASD) were compared using MANOVA on the five ITSP domains (auditory, visual, tactile, vestibular, oral sensory). A second MANOVA was performed on the four quadrant scores (low registration, sensory sensitivity, sensory seeking, and sensory avoiding) with alpha (i.e., overall type I error) set at 0.05. We also explored possible group differences in ITSP subscales and quadrants, applying a Bonferroni correction to set critical p-values for statistical significance (i.e., 0.05/5 = 0.01 and 0.05/4 = 0.0125, respectively).

Results

Participant Characteristics

A total of 31 LR and 60 HR infants (15 HR-ASD; 45 HR-N) had complete ITSP data at 24 months (mean age at assessment = 24.7 ± 1.1 months) and had 3-year diagnostic outcome data (mean age at assessment = 37.6 ± 1.8 months). One LR infant received an ASD diagnosis at age 36 months and was excluded from further analysis. This infant scored within the "Typical Performance" range for all ITSP domains and quadrants with the exception of visual processing and oral sensory processing. There were no gender or age differences between HR-ASD, HR-N, and LR groups (p's > 0.05) but, as anticipated, there were an group differences on MSEL scores (i.e., Early Learning Composite Standard Scores, Expressive Language T-Scores and Receptive Language T-Scores; p's < 0.001) and ADOS Severity scores (p's < 0.001). See Table 1 for details.

Sensory Processing

Groups differed significantly in auditory processing (e.g., "my child tries to escape from noisy environments;" $F(2,89) = 5.71$, $p = 0.005$), with the HR-ASD group scoring significantly higher than the HR-N and LR groups; the HR-ASD mean score fell within the "Probable Difference" range, in contrast to the HR-N and LR groups, whose scores fell within the "Typical Performance" range. No differences were detected in the visual, vestibular, tactile, or oral sensory processing domains (all p's > 0.05). See Table 2 for details.

Of the quadrant scores, a significant group difference was obtained only for low registration (e.g., does not notice sensory stimuli; $F(2,89) = 4.601$, $p = 0.013$). The mean score for the HR-ASD group was significantly higher than those of the HR-N and LR-groups, with the HR-ASD mean falling within the "Probable Difference (more than others)" range, and the HR-N and LR means within the "Typical Performance" range. The four quadrant scores are *not* specific to any particular sensory domain (i.e., auditory, visual, etc.) but rather reflect the toddlers' overall behaviours across the range of sensory inputs. See Table 2 for details.

As well, there was a non-significant trend towards group differences in the "Probable Difference" range for sensory-seeking behaviour (e.g., seeks out sensory stimulating environments) for HR-ASD, in comparison to the HR-N and LR infants ($F(2,89) = 3.242$, $p = 0.044$) after post hoc analysis. See Tables 3 and 4 for details on classification frequencies by domains and quadrants (respectively).

Discussion

The hypothesis of this study was only partially supported, as early parent-reported auditory processing differences and low registration (not specific to any sensory domain) were associated with a subsequent ASD diagnosis among 24-month-old HR infants. Within the ITSP, examples of atypical auditory processing, as reported by parents, include: 'my child tries to escape noisy environments' and 'my child enjoys making sounds with his/her mouth', whereas examples of low registration are: 'my child seems unaware of wet or dirty diapers' and 'my child bumps into things, seeming to not notice objects in the way' (Dunn 2002). These results suggest that parent-reported early sensory differences are potential risk markers of ASD, at least among HR toddlers. However, differences were found

Table 2 MANOVA results for sub-domains of sensory processing by HR-ASD, HR-N, and LR

	Mean (SD)			*F*-value (2,89)	*p* (α = 0.05)	Post-hoc (with Bonferroni correction)
	HR-ASD (a)	HR-N (b)	LR (c)			
Main effects of domains						
Auditory processing	2.53 (0.99)	3.47 (0.99)	3.16 (0.82)	5.711	**0.005**	a < b, c
Visual processing	3.47 (0.74)	3.53 (0.73)	3.45 (0.62)	0.188	0.829	ns
Tactile processing	3.20 (0.86)	3.47 (0.79)	3.29 (0.78)	0.743	0.479	ns
Vestibular processing	2.73 (0.80)	2.91 (0.76)	2.84 (0.69)	0.382	0.684	ns
Oral sensory processing	2.87 (0.92)	3.29 (0.73)	3.16 (0.69)	2.462	0.091	ns
Main effects of quadrants						
Low registration	2.33 (1.11)	2.96 (0.67)	2.90 (0.60)	4.601	**0.013**	a < b, c
Sensory sensitivity	2.93 (0.70)	3.07 (0.62)	2.87 (0.62)	0.918	0.403	ns
Sensory seeking	3.40 (0.91)	3.91 (0.82)	3.52 (0.68)	3.242	0.044	ns
Sensory avoiding	2.73 (0.80)	2.96 (0.70)	3.00 (0.73)	0.774	0.464	ns

The values in bold are significant, thus highlighting the need for further post-hoc analysis

Table 3 Auditory processing classifications by group

Auditory processing	HR-ASD (%)	HR-N (%)	LR (%)
Definite difference (more than others)	3 (21.43)	2 (4.44)	2 (6.54)
Probable difference (more than others)	3 (21.43)	3 (6.67)	0 (0)
Typical performance	6 (42.86)	19 (42.22)	22 (70.97)
Probable difference (less than others)	2 (14.28)	14 (31.11)	5 (16.13)
Definite difference (less than others)	0 (0)	7 (15.56)	2 (6.54)
Total	14 (100)	45 (100)	31 (100)

Table 4 Low registration classifications by group

Low registration	HR-ASD (%)	HR-N (%)	LR (%)
Definite difference (more than others)	5 (35.71)	3 (6.67)	2 (6.45)
Probable difference (more than others)	2 (14.29)	2 (4.44)	1 (3.22)
Typical performance	5 (35.71)	34 (75.56)	26 (74.29)
Probable difference (less than others)	2 (14.29)	6 (13.3)	2 (6.45)
Definite difference (less than others)	0 (0)	0 (0)	0 (0)
Total	14 (100)	45 (100)	31 (100)

Percentages are calculated based on the number of toddlers per group across classifications; Classification break down was *only* provided for domains and quadrants of significance

in only two ITSP domains. The gradient hypothesis regarding the expected sensory responses from HR-ASD, to HR-N and LR, was partially supported, with LR infants most often falling within the 'Typical Performance' range.

Previous research on early sensory development in ASD has focused on toddlers or children who have already received a diagnosis of ASD (Ben-Sasson et al. 2007, 2009; Kern et al. 2006, O'Donnell et al. 2012), with one prospective study of a small sample of high-risk infants in which diagnostic outcomes were not reported (Mulligan and White 2012). The present study builds on this work by examining whether parent-reported sensory processing symptoms varied within the HR group in relation to ASD outcome. An additional strength of the present study is the use of community controls, rather than normative data, as in a previous study (Mulligan and White 2012). The results presented here suggest that early differences in sensory processing may assist in differentiating between HR infants who later will and will not develop ASD.

In a previous case series of the first nine HR infants diagnosed with ASD from our prospective cohort, seven infants were observed clinically to demonstrate atypical sensory behaviour between 12 and 18 months, such as being distressed by noisy environments or the sound of running water, visual fixation on hands or objects, and mouthing and sniffing objects (Bryson et al. 2007). Difficulties with sensory processing (as indexed by parent report) have also been significantly associated with higher levels of problem behavior in preschool children with ASD (O'Donnell et al. 2012). Thus, atypical sensory processing in high-risk infants may reflect poor emotional regulation and atypical behavioral responses as part of a trajectory towards ASD diagnosis. Our primary hypothesis was partially supported, as auditory processing differences and low sensory registration were found for HR-ASD infants compared to HR-N and LR infants; however no significant differences in the domain or quadrant scores were found between HR-N and LR infants.

Other presumed sensory phenomena, such as texture aversions in eating, frequent rubbing of hands on novel textures, and visual fixation on objects or parts of the body, are sometimes reported to clinicians in young children with suspected ASD and/or detected in early home videos (Baranek 1999; Dickie et al. 2009). However, the ITSP did not detect such differences in the present study. Direct clinical observation of sensory behaviour and/or objective laboratory-based sensory measures (i.e., binaural psychoacoustic tests) could supplement parent reports to provide multiple perspectives on sensory processing in infants at high risk for ASD. In a related area of study for HR infants, temperament differences (i.e., difficulties in behaviour regulation compared to typically developing infants) on the basis of parent reports from 12 to 24 months were able to distinguish HR-ASD from HR-N/LR infants (Garon et al. 2009; Clifford et al. 2012) and HR-ASD from HR-N (del Rosario et al. 2014). Although temperament has not been explicitly explored in relationship to sensory processing in HR infants for ASD, it may provide a useful framework for sensory and regulatory differences reported early in development in ASD (Zwaigenbaum et al. 2012).

This study has several strengths that support the findings. The sample is larger than in previous reports (e.g., by Mulligan and White 2012), the HR group is stratified by 3-year diagnostic outcomes, and the data were gathered prospectively (i.e., without recall bias). However, there are also potential limitations. First, although larger than previous samples, in which early sensory processing has been assessed, our sample is still relatively small, and may fail to capture the heterogeneity in the population. Future studies could include examining sensory profiles for children with ASD on the basis of symptom levels and functional abilities to understand whether sensory differences are correlated with severity along these dimensions, although larger samples would be needed. As well, although we report significant group differences, there is overlap in auditory processing and registration across the HR-ASD and other groups at an individual level. Moreover, the underlying assumption of the ITSP is that a child's behavior is a reaction to sensory input from their surrounding environment. However, combination of the ITSP with objective observational sensory processing measurements, might prove more informative. As well, it is important to identify whether sensory processing differences exist between HR infants and infants with other developmental disabilities, especially to enhance the clinical utility of the ITSP. Based on the review by Rogers and Ozonoff (2005), good evidence does not presently exist to differentiate ASD from other developmental disability groups on the basis of sensory differences or profiles. Although this study contributes novel data regarding sensory differences predictive of ASD within a HR cohort, the degree to which these findings generalize to children who do not have a family history of ASD remains to be examined. Finally, it could be postulated that second-time parents (which all HR infants were) would be more astute regarding their children's development, having their experiences with their first-children as a basis for comparison. As such, our parent responses may be more reliable than first-time parents.

Nevertheless, our findings emphasize the importance of parent reports of sensory difficulties at 24 months, particularly in the auditory domain, as a component of prospective monitoring of infants at high-risk of ASD. The ITSP can play a role in exploring parent-observed behavioral responses to sensory input, such as differences with auditory processing or lower registration, as part of this monitoring process.

Acknowledgments This study was supported by the Canadian Institutes of Health Research, Autism Speaks, Alberta Innovates – Health Solutions, Stollery Children's Hospital Foundation, NeuroDevNet and Autism Research Training program. The authors thank all the families who participated in the *Canadian Infant Siblings Study*. The authors also acknowledge the members of the *Canadian Infant Siblings Study Team*: Sanne Jilderda, Jonathan Leef, Bonnie MacKinnon, Stacey MacWilliam, Theresa McCormick, Mayank Rehani, Ellen Robertson, Jana Roberto, Ann Thompson and Carol Wilson.

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