Sensory Features of Toddlers at Risk for Autism Spectrum Disorder

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MeSH TERMS
- autism spectrum disorder
- child development
- early diagnosis
- risk factors
- sensation
- sensation disorders

OBJECTIVE. We observed sensory features in toddlers ages 12–24 mo with risk factors for autism spectrum disorder (ASD) and explored their relationship to general development and early signs of ASD.

METHOD. Participants (N = 46) included toddlers with higher risk for ASD. All participants were administered standardized assessments of sensory features, early signs of ASD, and general development at a single study visit.

RESULTS. Sensory features in toddlers were characterized as either adaptive or reactive. Toddlers with more difficulties in oral sensory processing displayed more early signs of ASD. Typical oral and auditory processing were associated with higher cognitive function, and toddlers with fewer sensory features overall had more mature language skills.

CONCLUSION. Specific sensory features were associated with both early signs of ASD and less mature general development. Replication of this preliminary study is required.

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Sensory features are now included as one aspect of restricted and repetitive behaviors included in the diagnostic criteria for autism spectrum disorder (ASD; American Psychiatric Association [APA], 2013). *Sensory features* are behavioral responses to sensory stimuli characterized by hyperreactivity, hyporeactivity, and unusual sensory interests (APA, 2013). Important differences in sensory features have been identified between older children diagnosed with ASD and control groups. Previous studies have highlighted differences in the auditory, vestibular, proprioceptive, tactile, and oral domains (Ludlow et al., 2014; Tomchek & Dunn, 2007). Little is known, however, about the emergence of sensory features in young children with risk factors for ASD.

Previous studies in which researchers examined sensory features in young children at risk for ASD relied on parent recall or retrospective video analysis (Baranek, 1999; Clifford, Young, & Williamson, 2007; Watson et al., 2007). These preliminary studies suggested that the presence of unusual sensory behaviors—such as visual fascinations, excessive mouthing, and aversion to social touch—can distinguish infants later diagnosed with ASD from typically developing peers (Baranek, David, Poe, Stone, & Watson, 2006). These studies are limited by the potential for recall bias and at times the absence of an appropriate comparison group (Reznick, Baranek, Reavis, Watson, & Crais, 2007). No studies have attempted to examine specific patterns of sensory features in early childhood that may be associated with ASD risk.

Literature to date has suggested that sensory features are pervasive in ASD, affecting up to 92% of children (Baranek, Boyd, Poe, David, & Watson, 2007; Ben-Sasson et al., 2009). In two recent studies, researchers identified patterns of sensory features, or *subtypes,* in populations with ASD (Ausderau et al., 2014;

Lane, Molloy, & Bishop, 2014). Lane and colleagues (Lane, Dennis, & Geraghty, 2011; Lane et al., 2014; Lane, Young, Baker, & Angley, 2010) observed four distinct sensory subtypes in ASD that differ in the severity and focus of sensory symptoms. Specifically, the authors proposed that difficulties in sensory reactivity and multisensory integration underpin the variation in the subtypes. *Sensory reactivity* refers to the ability to modulate the intensity of a response when exposed to a given stimulus. In contrast, *multisensory integration* is the process of perceiving and interpreting stimuli from multiple sensory modalities presented concurrently (Baranek, Little, Parham, Ausderau, & Sabatos-DeVito, 2014). In the sensory classification system proposed by Lane et al. (2014), children with ASD may present with (1) no sensory symptoms (sensory adaptive), (2) symptoms of sensory reactivity (hyper- or hyporeactivity) only, (3) symptoms of multisensory integration difficulties only, or (4) symptoms of both sensory reactivity and multisensory integration difficulties.

Previous research has provided strong evidence for a positive relationship between symptoms of ASD and sensory features in multiple sensory domains (Schaaf & Lane, 2015); however, little prediagnostic research exists. Sensory features can influence a range of core ASD behaviors in young children, including daily living skills, restricted and repetitive behaviors, and social communication skills (Hazen, Stornelli, O'Rourke, Koesterer, & McDougle, 2014; Jasmin et al., 2009). Studies have shown that children with ASD who exhibit hyper- or hyporeactivity to sensory stimuli also display increased repetitive behaviors, such as stereotypies, need for sameness, and self-injurious behaviors (Boyd et al., 2010; Duerden et al., 2012; Gabriels et al., 2008). Additionally, issues with social communication—such as failure to respond during social contexts, avoidance, and reduced orienting to salient social stimuli—have also been attributed to sensory hyporeactivity in children with ASD (Baranek et al., 2006; Ben-Sasson et al., 2007; Watson et al., 2011).

In our study, we aimed to extend understanding of the relationship between sensory features and the emergence of early ASD signs. First, we identify and describe sensory subtypes in toddlers ages 12–24 mo with and without risk for ASD. We hypothesized that toddlers would display (1) adaptive sensory function, (2) sensory features characterized by hyper- or hyporeactivity, (3) sensory features characterized by difficulties in multisensory integration, or (4) sensory features characterized by difficulties in both sensory reactivity and multisensory integration. Second, we explore the relationship among sensory features, the presence of early signs of ASD, and general development in toddlers ages 12–24 mo. We hypothesized that toddlers showing elevated early signs of ASD or developmental delay would also display sensory features.

Method

Design

In this one-sample, descriptive study, we evaluated ASD risk status, general development, and sensory features of a single cohort of toddlers at one time point. There were no comparison or control groups.

Participants

Forty-six toddlers participated in this study. We used purposive sampling to target toddlers with higher risk for ASD during recruitment, such as siblings of children diagnosed with ASD and those with a history of premature birth. Recent research has shown lower gestational age to be a possible risk factor for ASD, with higher prevalence rates among children born extremely preterm (<28 wk) than among full-term peers (Cassimos, Syriopoulou-Delli, Tripsianis, & Tsikoulas, 2015; Guy et al., 2015; Kuzniewicz et al., 2014). Thus, premature infants were included in this study. Participants were recruited from child care and medical centers as well as private therapy practices in a large urban center in the midwestern United States. Parents provided informed consent before their child participated in the study. The sample consisted of 2 siblings of children with ASD, 4 toddlers born prematurely before 37 wk gestation, and 40 toddlers born full term.

Toddlers were included if they were between 12 and 24 mo old and had intact hearing (assessed by parent report of newborn hearing screening). Toddlers diagnosed with a non-ASD condition—such as spina bifida, cerebral palsy, or Down syndrome—were excluded to reduce the risk of confounding because of other comorbidities.

Procedure

The institutional review boards at the Ohio State University, the Nationwide Children's Hospital, and the University of Newcastle Australia Human Research Ethics Committee approved this study (H-2014-0017). The third author (Harpster) administered clinical measures to examine sensory features, ASD risk, and general development in a single visit at the research laboratory of the second author (Lane).

Instrumentation

Parent-reported sensory features were assessed with the Infant–Toddler Sensory Profile (ITSP; Dunn, 2002).

The ITSP is a norm-referenced, standardized parent-report questionnaire used to examine a toddler's reactions to sensory input in his or her typical environments. The scale consists of 48 items rated on a 5-point Likert scale ranging from 1 (*almost always*) to 5 (*almost never*). Sensory features and behaviors are scored along a continuum of hypo- and hyperreactive behavioral patterns as well as typical behavioral responses in visual, auditory, tactile, vestibular, and oral sensory processing domains. ITSP z scores were calculated with normative data from the ITSP user's manual. Scores falling within 1 standard deviation of the mean are categorized as typical performance. Lower scores on the ITSP indicate that a child falls within the less-than-others range, which equates to 1 or 2 standard deviations below the mean. We interpreted z scores falling outside 1 or 2 standard deviations of the mean as denoting hyperreactive responses to sensory stimuli, indicating clinically important issues in sensory processing.

The ITSP has previously been used to examine sensory features in high-risk infants (Germani et al., 2014). Previous research has highlighted adequate test–retest reliability, internal consistency, and content and concurrent (criterion) validity of the ITSP (Dunn & Daniels, 2002; Eeles et al., 2013).

The Autism Detection in Early Childhood (ADEC; Young, 2007) was used to determine the presence or absence of early signs of ASD at 12–24 mo. The ADEC is a standardized observational measure consisting of 16 items assessing preverbal behaviors, such as response to name, functional play, eye contact, and imitation. Children score between 0 and 2 on each item, with a higher total score indicating an increased presence of early ASD signs. Strong psychometric properties have been established for the measure, including high internal consistency, interrater reliability, test–retest reliability, sensitivity (1.0), and specificity (0.74–0.90), as well as construct and concurrent validity of the ADEC as an early ASD screening tool (Nah, Young, Brewer, & Berlingeri, 2014).

General development was assessed with the Bayley Scales of Infant and Toddler Development—Third Edition (Bayley–III; Bayley, 2006). The Bayley–III is a standardized, norm-referenced observational tool for evaluating general development in young children ages 1–42 mo. This tool is used to examine cognition, expressive and receptive communication, and fine and gross motor skills. A scaled score between 8 and 12 indicates skills within typical performance. Previous research has highlighted adequate content validity, construct validity, and internal consistency of the Bayley–III (Bayley, 2006; Spittle, Doyle, & Boyd, 2008).

Analysis

All statistical analyses were conducted with IBM SPSS Statistics (Version 21.0; IBM Corp., Armonk, NY) and R (Version 2.15; R Foundation for Statistical Computing, Vienna, Austria). Descriptive statistics were used to establish the means and standard deviations for all participants in relation to each measure.

Model-based cluster analysis (Stahl & Sallis, 2012) was applied to ITSP z scores to address the first aim. This type of analysis is used to identify homogeneous subgroups in populations demonstrating similar characteristics. In model-based cluster analysis, one uses the Bayesian information criterion to determine the best cluster model by comparing the fit of the data with a range of possible models. This method of analysis is appropriate when attempting to establish how a specific construct—in this case, sensory features—varies across a population (Zhong & Ghosh, 2003). Diagnostic homogeneity is not a prerequisite for the use of this analysis approach; instead, it is assumed that the observed data arise from a population containing several subpopulations (Raftery & Dean, 2006). The purpose of our study was to determine whether systematic variations in sensory features in a sample of toddlers with and without risk factors for ASD conform to the variations previously reported in older children with ASD (Lane et al., 2014) and whether these patterns of variations are associated with risk for ASD.

We used the Pearson product–moment correlation coefficient (Pearson's r) to address the second aim. Moreover, we examined differences between sensory subtypes (identified in the "Aim 1" section) on the ADEC and Bayley–III using one-way analysis of variance (ANOVA).

Results

Descriptive statistics for the total sample on each instrument are displayed in Table 1.

Aim 1: Sensory Subtypes

Examination of the Bayesian information criterion values after model-based cluster analysis revealed a four-cluster solution as the most parsimonious (see Figure 1). Scatterplots of pairwise comparisons of each ITSP domain suggest that the clearest differentiation among clusters occurs when contrasting visual and auditory domains (see Figure 2).

Figure 2 shows two primary clusters (Cluster 2, $n = 19$; Cluster 3, $n = 18$) and two secondary clusters (Cluster 1, $n = 4$; Cluster 4, $n = 5$). One-way ANOVA was used to identify differences among clusters in all ITSP domains

Table 1. Mean Scores (and Standard Deviations) for ITSP Domains, ADEC Total Score, and Bayley–III Categories by Cluster

Variable	Cluster 1, *M* (*SD*)	Cluster 2, *M* (*SD*)	Cluster 3, *M* (*SD*)	Cluster 4, *M* (*SD*)
Initial *N*	4	19	18	5
Initial *M* age, mo	19.5	17.3	18.5	16.6
ITSP, *M* raw scores				
Auditory	37.75 (0.96)	34.95 (3.99)	41.78 (2.58)	40.20 (1.30)
Visual	20.25 (0.96)	21.58 (2.69)	26.33 (3.03)	22.00 (0.00)
Tactile	58.25 (2.06)	51.05 (5.24)	60.11 (3.53)	55.40 (2.19)
Vestibular	21.50 (0.58)	17.11 (2.03)	21.22 (2.10)	18.20 (0.84)
Oral	29.75 (0.50)	26.16 (4.34)	28.22 (2.92)	29.60 (1.14)
ADEC, total score	6.00 (4.40)	6.47 (3.24)	5.33 (3.45)	5.20 (2.39)
Bayley–III, *M* scaled score				
Cognitive	11.00 (2.31)	11.53 (2.01)	12.22 (2.58)	14.00 (3.00)
Receptive communication	9.50 (3.70)	9.58 (2.83)	12.29 (2.89)	13.40 (2.61)
Expressive communication	8.25 (3.74)	8.47 (2.41)	11.56 (1.92)	12.80 (3.77)
Fine motor	10.75 (1.26)	11.58 (2.14)	12.00 (2.45)	13.40 (3.36)
Gross motor	10.75 (3.77)	9.42 (2.09)	9.94 (2.18)	13.40 (3.51)

Note. For the total sample, initial $N = 46$ (28 boys, 18 girls), and the initial *M* age = 17.9 mo. ADEC = Autism Detection in Early Childhood; Bayley–III = Bayley Scales of Infant and Toddler Development—Third Edition; ITSP = Infant–Toddler Sensory Profile; *M* = mean; *SD* = standard deviation.

except for oral sensory processing (see Table 2). Tukey's post hoc analyses revealed that Cluster 2 differed significantly from Cluster 1 in the vestibular domain ($p = .001$) and from Cluster 3 in the auditory ($p = .001$), vestibular ($p = .001$), visual ($p = .001$), and tactile ($p = .001$) domains. Cluster 2 also differed significantly from

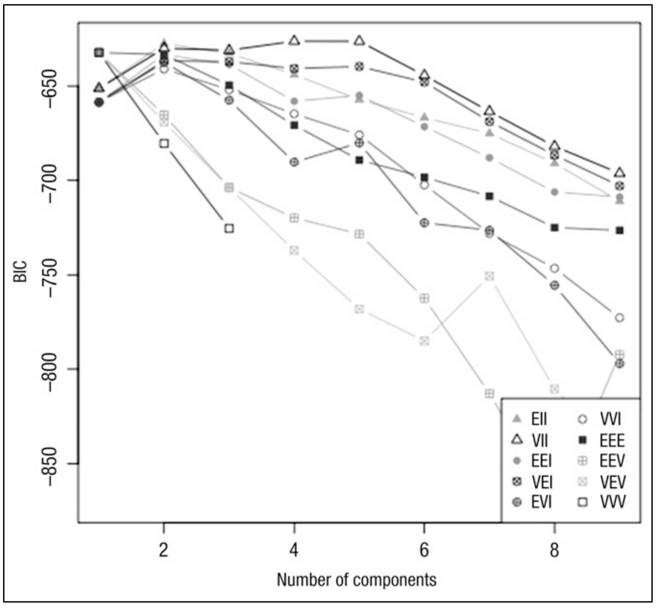

Figure 1. Bayesian information criterion (BIC) values for each model considered for the model-based cluster analysis, with each model being represented by a different symbol. The BIC peaks at four components, suggesting that this value is the most parsimonious solution for the analysis.

Note. EEE = ellipsoidal, equal volume, equal shape, and equal orientation; EEI = diagonal, equal volume, equal shape; EEV = ellipsoidal, equal volume, equal shape; EII = spherical, equal volume; EVI = diagonal, equal volume, varying shape; VEI = diagonal, varying volume, equal shape; VEV = ellipsoidal, equal shape; VII = spherical, unequal volume; VVI = diagonal, varying volume, varying shape; VVV = ellipsoidal, varying volume, varying shape, varying orientation.

Cluster 4 in the auditory domain ($p = .004$). In these sensory domains, Cluster 2 mean ITSP *z* scores suggested sensory hyperreactivity, established by the ITSP because general items scoring below the mean (0) are suggestive of hyperreactivity. In contrast, Clusters 1, 3, and 4 had scores within 1 standard deviation of the normative mean in both visual and auditory domains, suggesting typical sensory function across these domains. On the basis of previous subtyping research (Lane et al., 2014) and the results of our analysis, we suggest that the four clusters identified in this sample are well represented by two broad subtypes:

1. *Sensory reactive* (Cluster 2)—All scores fall below the normative mean with subfoci in auditory and vestibular domains.
2. *Sensory adaptive* (Clusters 1, 3, and 4)—Most scores fall within 1 standard deviation of the normative mean.

Aim 2: Sensory Features, Early Signs of Autism Spectrum Disorder, and General Development at 12–24 Mo

A moderate, but significant, negative correlation between oral sensory processing and ADEC score ($r = -.32$, $p = .03$) was observed. This finding suggests that participants with more oral sensory processing difficulties also displayed higher risk factors for ASD. There were no other significant correlations between the ITSP and ADEC. Oral sensory and auditory processing were, however, moderately and positively correlated with cognitive scores ($r = .40$, $p = .01$, and $r = .31$, $p = .03$, respectively), suggesting that higher cognitive function is associated with more typical sensory processing in these domains.

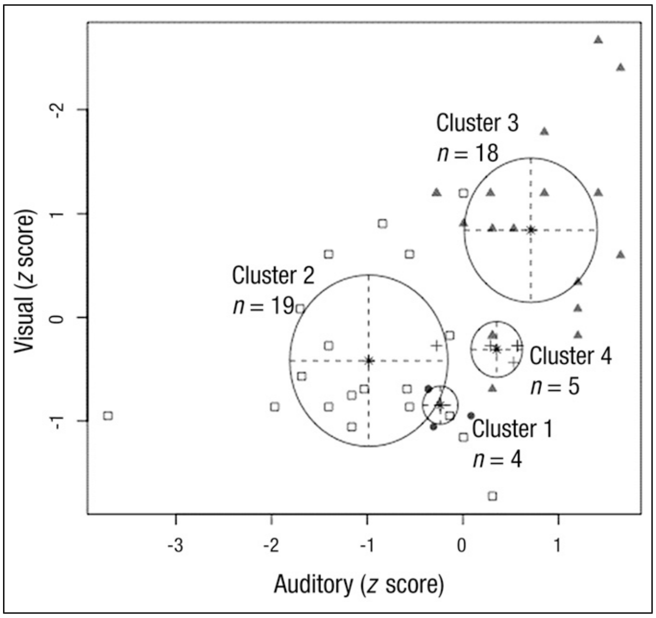

Figure 2. Cluster solution as a function of Infant–Toddler Sensory Profile auditory and visual domains. A value of 0 represents normative mean for performance on the sensory domains (i.e., typical performance). Values greater than and less than 0 indicate the number of standard deviations away from the mean. Values between −1 and 1 indicate typical performance. Values greater than 1 indicate significant issues with hyporeactivity. Values less than −1 indicate significant issues with hyperreactivity.

Significant, moderate correlations were also observed among auditory ($r = .55$, $p = .001$), visual ($r = .31$, $p = .04$), tactile ($r = .32$, $p = .03$), and oral ($r = .34$, $p = .02$) sensory domains as well as receptive communication scores. Similar correlations were found among expressive communication scores and auditory ($r = .49$, $p = .001$), visual ($r = .41$, $p = .01$), tactile ($r = .41$, $p = .01$), vestibular ($r = .32$, $p = .03$), and oral ($r = .35$, $p = .02$) sensory domains. These results indicated that more mature language abilities were associated with fewer sensory features. No associations were observed among sensory features, fine motor scores, and gross motor scores.

When examined by sensory subtype, one-way ANOVA revealed significant differences between the sensory reactive and sensory adaptive subtypes in relation to expressive communication ($p = .001$) and receptive communication ($p = .01$). Participants with sensory reactivity displayed less mature language abilities. No significant differences were found among subtypes on ADEC score at 12–24 mo or on cognitive or motor scores.

Discussion

This study revealed distinct prediagnostic sensory subtypes in toddlers with and without ASD risk. The findings provide further preliminary evidence that early sensory features are present in toddlers with heightened risk for ASD and that these features are associated with discrete ASD-related symptoms. Specifically, we observed and characterized two sensory subtypes: (1) sensory reactive (with hyperreactivity in auditory and vestibular domains) and (2) sensory adaptive (generally with typical sensory function).

In this study, we did not find a specific subtype characterized by difficulties with multisensory integration, which has previously been established (Lane et al., 2014). This result may have been due to the content of the ITSP, which is focused on sensory reactivity (Dunn & Daniels, 2002). Further research is needed to clarify the nature of multisensory integration difficulties that may exist in early childhood.

Toddlers in our study with less mature oral sensory processing abilities were more likely to display higher ASD risk. In previous studies, researchers have reported a high presence of oral sensory features in children at risk for ASD, which has been further associated with food selectivity and picky eating (Cermak, Curtin, & Bandini, 2014). Toddlers in our study classified as sensory reactive demonstrated the lowest average oral sensory processing scores, which supported this trend. However, this trend was not considerable, likely because of our small sample size and because all toddlers in this sample presented with elevated oral sensory processing difficulties. Additional investigation is warranted with a sample displaying a greater range of oral sensory processing abilities.

Important differences were observed between the sensory reactive and sensory adaptive subtypes in early language development. Compared with their peers, toddlers in the sensory reactive subtype displayed less mature language abilities at 12–24 mo. This relationship has not previously been reported, although links between sensory

Table 2. Mean ITSP Domain *z* Scores (and Standard Deviations) by Cluster With ANOVA Results

ITSP Domain	Cluster 1, *z* (*SD*)	Cluster 2, *z* (*SD*)	Cluster 3, *z* (*SD*)	Cluster 4, *z* (*SD*)	*p*
Auditory	−0.24 (0.22)	−1.00 (0.93)	0.72 (0.63)	0.34 (0.36)	.001
Visual	−0.85 (0.18)	−0.43 (0.78)	0.85 (0.90)	−0.31 (0.07)	.001
Tactile	0.53 (0.30)	−0.57 (0.82)	0.83 (0.56)	0.10 (0.35)	.001
Vestibular	0.54 (0.16)	−1.17 (0.82)	0.39 (0.77)	−0.80 (0.34)	.001
Oral	−1.09 (0.16)	−1.81 (0.87)	−1.46 (0.63)	−1.10 (0.28)	.100

Note. ANOVA = analysis of variance; ITSP = Infant–Toddler Sensory Profile; *SD* = standard deviation.

features and impaired social communication skills in young children with risk factors for ASD have been reported (Mitchell et al., 2006; Watson et al., 2011). However, our results should be interpreted cautiously because mean scores for each subtype did not fall outside the range for typical function on the communication scales. As a group, participants in the sensory reactive subtype did not present with a true language delay; however, they did present with a pattern of less mature language development.

Overall, the small sample size and cross-sectional design of this study limit the conclusions that can be drawn. In future studies, researchers should augment the parent-report measures used in this study with more objective measures of sensory features. Although trends can be detected in the data, participants' scores did not vary greatly across the sample, and many children performed within a typical range of function. Future research with larger samples and higher variance among participants is needed to confirm our results.

Implications for Occupational Therapy Practice

In this study, we identified that a subset of toddlers with risk factors for ASD exhibited prediagnostic sensory reactivity. We also highlighted a potential relationship among sensory features, cognition, language development, and ASD signs at 12–24 mo. Overall, the results of this study

- Have provided a basis on which to conduct further studies with larger datasets that can more precisely delineate which sensory features are critical in the emergence of ASD in later childhood,
- Can be used to guide the observations of occupational therapists working in early intervention, and
- Have shown that the ITSP is a useful tool in identifying specific patterns of sensory features that may be relevant to the emergence of ASD symptoms.

Conclusion

The findings from this study add to previous literature by providing an understanding of sensory-based subtypes in a prediagnostic ASD sample. The prevalence of two distinct subtypes, as characterized by reactive and adaptive sensory features, was observed for young children ages 12–24 mo. Overall, the young children displaying both signs of sensory reactivity and less mature language development may be well served by further, comprehensive evaluation to rule out a diagnosis of ASD. Further research is required to examine the functional implications of the sensory subtypes established in this study to guide intervention procedures for children at risk of ASD who exhibit atypical sensory processing. ▲

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