

A Population-Based Twin Study of Parentally Reported Tactile and Auditory Defensiveness in Young Children

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Some adults and children exhibit defensive behaviors to tactile or auditory stimulation. These symptoms occur not only in subsets of children with ADHD, autism, and Fragile X syndrome, but also in the apparent absence of accompanying disorders. Relatively little research explores the correlates and antecedents of sensory defensiveness. Using a population-based sample of 1,394 toddler-aged twins, mothers reported on tactile and auditory defensiveness, temperament, and behavior problems. The incidence of defensive symptoms was widely distributed, with some accumulation of cases in the extreme range. Girls were overrepresented in the extreme tactile defensiveness group. Both auditory and tactile defensiveness were modestly associated with fearful temperament and anxiety, but they were relatively distinct from other common dimensions of childhood behavioral dysfunction. Twin correlations for the full range of scores and concordance rates for the extremes suggested moderate genetic influences, with some indication that the tactile domain might be more heritable than the auditory domain.

KEY WORDS: sensory defensiveness; twins; temperament; genetics; anxiety.

INTRODUCTION

Some children fuss about stiff new clothes, turtleneck sweaters, and labels sewn inside collars. These children may be bothered by the seams of socks, dislike being touched lightly on the face, protest vigorously to fingernail trimming and tooth brushing, and demonstrate other behaviors that fall under the rubric of “tactile defensiveness.” A comparable set of symptoms, such as highly aversive reactions to vacuum cleaners and sirens, might indicate “auditory defensiveness.” Health professionals regard these sensitivities in vastly different ways. On the one hand, some occupational therapists include tactile and auditory defensiveness as two among several hyper- and hypo-sensitivities that define a clinical entity called sensory modulation disorder or sensory integration dysfunction (Dunn, 2001). Whether or not

sensory modulation disorder has the features that should lead the broader field to recognize it as a discrete clinical entity is debatable. However, at least some symptoms that seemingly reflect reactions to sensory stimulation occur in subsets of children with attention deficit hyperactivity disorder (ADHD; Mangeot et al., 2001), autism (Kientz & Dunn, 1997), and Fragile X syndrome (Cohen, 1995). The same behaviors also appear in children without any formal diagnosis. On the other hand, some professionals minimize the significance of putative sensory defensiveness. They regard these behaviors much like mild clumsiness or mild shyness, as problems that typically do not rise to a level of clinical significance, or that are secondary to other more basic dysfunctions.

Thus, we were motivated to explore sensory modulation difficulties, without any strong bias regarding whether they constitute a disorder, but with the assumption that the relevant behaviors are real—sometimes debilitating—and deserve systematic study. Rather than assuming the existence of sensory modulation disorder, we use the term, “sensory defensiveness,” and define it operationally as hedonically negative behavioral responses indicating

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withdrawal or protest that appear to be unusual reactions to noxious levels of sensation. Our study involves parental report of tactile and auditory reactions of twin children, who were systematically ascertained from statewide birth records. The use of twins allows us to estimate genetic influence on differences in tactile and auditory defensiveness; findings of genetic influence would strengthen the case for the reality of the phenomena. The use of a behavioral symptom inventory allows us to place tactile and auditory defensiveness within a larger clinical context.

Literature Review

Three domains of prior research constitute background for this study. First is the measurement tradition in the field, which is the use of parental report questionnaires. The second domain relates sensory defensiveness to other aspects of functioning, particularly to behavioral disorders. The final domain concerns the plausibility of genetic influences on sensory defensiveness.

Measurement

Assessing sensory modulation by observation of a child in several situations designed to elicit negative behavioral responses would present obvious difficulties for a research study. Although standardized observations are needed, researchers have instead relied on parent-report questionnaires. Parental report measures of children's sensory defensiveness are potentially subject to various biases, the strength and seriousness of which have been analyzed in other domains, such as temperament (Goldsmith & Hewitt, 2003; Kagan, 1994, 1998; Mangelsdorf, Schoppe & Buur, 2000; Rothbart & Bates, 1998; Rothbart & Goldsmith, 1985). In the sensory defensiveness area, the amount of evidence bearing on the validity of parental report is limited and sometimes indirect. However, questionnaire measures of sensory defensiveness have become increasingly sophisticated and well-standardized. Their low cost and predominance in sensory defensiveness research justifies their use in initial genetic investigations.

The history of parent-report questionnaires of sensory defensiveness in children dates back over 25 years (Wilbarger, 1977). For instance, Provost and Oetter (1993) developed a 136-item questionnaire to assess several sensorimotor responses in infants and toddlers, and substantial psychometric work has been devoted to research versions of the Evaluation of Sensory Processing (ESP) instrument (Johnson-Ecker & Parham, 2000). Perhaps the most widely used instrument is the 125-item

Sensory Profile (Dunn, 1994), which groups items according to sensory domain. The Short Sensory Profile (Dunn, 1999) assesses a broad range of sensory issues in children 3–10 years, and a version for infants and toddlers is also available (Dunn, 2002). This brief measure has a coherent factor structure and high internal reliability (McIntosh, Miller, Shyu, & Dunn, 1999). Another caregiver-report questionnaire for young children is the Sensory Supplement Questionnaire (Baranek, 1999b), which has also shown promising psychometric properties. Our study was designed before the latest of these questionnaires were available; however, the item set that we developed shared item content with these newer instruments.

Association of Sensory Defensiveness with Other Childhood Behavior Problem Domains

Although work examining sensory defensiveness in children with clinical disorders is still in its early stages, some consistent results have emerged. First, individuals with certain disorders exhibit distinct patterns of physiological arousal to sensory stimulation. Second, clinical groups can be distinguished using both parent-report and physiological measures of sensory defensiveness. Third, anxiety appears to be a core feature of the disorders that co-occur with sensory defensiveness.

Children with ADHD, compared with typically developing children, tend to display greater difficulties in sensory modulation on both physiological and parent-report measures (Dunn & Bennett, 2002; Mangeot et al., 2001). Mangeot et al. (2001) found more variability in sensory defensiveness within a group of children with ADHD than within a comparison group. They interpreted this as evidence that children with ADHD are not a homogeneous group and suggested the existence of two ADHD subgroups distinguished by presence/absence of sensory modulation dysfunction.

Although sensory dysfunction has been considered a core deficit in autism, questions concerning the specificity of these sensory symptoms, the developmental pattern, and their relation to the severity of other symptoms still remain. Rogers, Hepburn, and Wehner (2003) found significantly elevated levels of sensory symptoms in children with autism compared with both typically developing children and children with delayed development. Sensory symptoms were elevated on tactile, gustatory, olfactory, and auditory filtering and were independent of symptoms in the social-communication domain. Furthermore, sensory symptoms and various kinds of repetitive and restrictive behaviors were already present in children with autism by 2.5 years of age. However, Lord (1995) has

suggested that sensory symptoms might increase as children develop through the preschool period. Both Kientz and Dunn (1997) and VerMaas Lee (1999) found substantial differences in sensory defensiveness between children with autism and typically developing groups on parental report questionnaires. In extensions of their main analyses of group mean differences, Kientz and Dunn observed no relationship between autism severity and sensory symptom severity. VerMaas Lee noted that auditory and proprioceptive items most strongly differentiated autistic and control groups but that differences were apparent in all sensory domains examined. In another study, Dunn, Myles, and Orr (2002) found a clear difference in the sensory defensiveness patterns of children with Asperger syndrome versus children without disabilities. In summary, these and other studies demonstrate that sensory defensiveness is much more common in children with autism than in typically developing children. A reasonable start has been made in investigating the nuances of this association.

Rogers et al. (2003) observed increased levels of sensory symptoms in children with Fragile X syndrome. Their findings indicated that, although children with autism had much higher levels of repetitive behavior than children with Fragile X syndrome, sensory behaviors were comparable in these two groups. Children with comorbid Fragile X syndrome and autism had the most severe sensory symptoms, but this observation based on seven children needs to be replicated. Bregman, Leckman, and Ort (1988) suggested that gaze aversion in Fragile X syndrome is related to anxiety. Belser and Sudhalter (1995) found that boys with Fragile X syndrome were more physiologically aroused by eye contact than were boys with Down Syndrome, suggesting links among poor eye contact, hyperarousal, and anxiety. A more recent study replicated and expanded Belser and Sudhalter's work by showing that boys with Fragile X syndrome have increased electrodermal reactivity (EDR) and less habituation to sensory stimuli and arousing situations (Miller et al., 1999).

A small literature suggests that individual differences in auditory sensitivity are associated with individual differences in socially withdrawn behavior. Aron and Aron (1997) reviewed the literature relating sensory processing and introversion and presented a model of sensory processing sensitivity. Socially withdrawn or introverted adults showed greater physiological reactivity to auditory stimulation as evidenced by larger auditory evoked response potentials (ERP; Doucet & Stelmack, 2000; Stelmack, Achorn, & Michaud, 1977). More recent work also suggests that social withdrawal in children may be related to individual differences in auditory processing. Specifically, socially withdrawn children show reduced

mismatch negativity (MMN) ERP amplitude and delayed latency to an auditory discrimination task (Bar-Haim, Marshall, Fox, Schorr, & Gordon-Salant, 2003). These researchers pointed out that a reduced MMN response to auditory discrimination also occurs in schizophrenia and depression. Other research has shown that individuals with schizophrenia and bipolar disorder score high on sensation avoiding, with individuals with schizophrenia also tending to miss available sensory stimuli (Brown, Cromwell, Filion, Dunn, & Tollefson, 2002).

In summary, sensory defensiveness apparently plays a role in various behavior problem domains, of which anxiety seems to be a feature. However, this could be due to the ubiquitous nature of anxiety. No study has focused directly on the relationship between anxiety and sensory defensiveness.

The Plausibility of Genetic Effects on Sensory Defensiveness

Clearly, individuals vary tremendously in how they respond to sensory stimuli. Some are sensation seekers whereas others avoid certain kinds of sensory experience as much as possible (Dunn, 1997). Such variability is a prerequisite for demonstrating genetic underpinnings, which have not been investigated very extensively. Goldsmith, Buss, and Lemery (1997) estimated twin similarity of a perceptual sensitivity scale from the Children's Behavior Questionnaire (Rothbart & Ahadi, 1994; Rothbart, Ahadi, Hershey, & Fisher, 2001). Identical and fraternal twin correlations were .58 and .37, respectively. Two other twin studies utilized a scale intended to measure the "ability to react to sensory stimuli of low stimulative value." Zawadzki, Strelau, Włodzimierz, Riemann, and Angleitner (2001) obtained self-reported temperament ratings for sensory sensitivity on two samples of adult twins. Genetic factors accounted for 38% of the individual variation in the first sample and 47% in the second sample. Nonshared environmental factors accounted for the remaining variance. This same pattern was reported by Oniszczenko (2002), who administered a child-appropriate version of the same questionnaire to parents of twins aged 6–11.

Both Strelau's sensory sensitivity scale and Rothbart's perceptual sensitivity scale refer to an individual's ability to detect slight sensations, rather than the negative reaction to sensory experiences typical of children with sensory modulation problems. Therefore, these three twin studies did not address the plausibility of genetic effects on sensory defensiveness per se. However, according to Strelau (1998), sensory sensitivity forms one dimension of

an individual's reactivity level. Highly sensitive individuals are expected to react with greater intensity to relatively low levels of stimulation. On the basis of this theory, we might expect that disturbances in behavior arise when one's sensitivity to sensory experiences does not match the stimulative value of those experiences (Strelau, 1998).

To our knowledge, ours is the first study of the genetic structure of extreme sensory defensiveness. However, as mentioned earlier, different sensory defensiveness patterns distinguish children with the single-gene Fragile X syndrome as well as highly heritable autism. In addition, sensory defensiveness has been closely linked to temperamental threshold of responsiveness, irritability, and fearfulness (Cohn, Miller, & Tickle-Degnen, 1999; Dunn, 2001), which are all influenced by genetic factors (Cyphers, Phillips, & Fulker, 1990). Therefore, it is reasonable to suspect that genetic factors may contribute to the individual variation in sensory defensiveness.

Not all findings suggest that sensory defensiveness behaviors are inherited, however. Environment, specifically prenatal environment, might also play a strong role in individual differences in sensory defensiveness. Schneider (2004) has shown that primates prenatally exposed to alcohol, lead, or high cortisol levels exhibited less habituation to tactile stimulation (stroking with a feather, cotton ball, or stiff brush) than did control animals. Exposed animals were also more likely to withdraw or exhibit other negative emotions than were control animals.

Exploring the genetic structure of sensory defensiveness, especially among extreme cases, will help resolve some of the issues surrounding these behaviors. Some clinical research suggests that sensory defensiveness is a distinct phenomenon rather than an extreme form of typical behavior (Lane, 2002). Genetic analyses allow us to determine if sensory defensiveness symptoms are more susceptible to genetic influences in the extreme cases than in typically developing children. Also, children may respond differently to sensory experiences in different modalities. Some research suggests that a general sensory modulation disorder underlies symptoms of both tactile and auditory defensiveness (McIntosh, Miller, Shyu, & Hagerman, 1999), but the specificity issue remains open. Again, knowing if one type of sensory defensiveness shares genetic variance with another could add to our understanding of the relationship between the two sensory domains.

Hypotheses and Research Questions

Given the limitations in the literature just reviewed, this study is necessarily partially exploratory. On an a priori basis, we cannot predict whether tactile and audi-

tory defensiveness scores will be distributed continuously throughout the population or, alternatively, reveal a set of children whose scores are separated from the bulk of the distribution. Thus, the distributional issue must be taken as a research question without a clear hypothesis. We hypothesize that defensiveness in the tactile and auditory domains can be usefully differentiated. We also hypothesize that high tactile and auditory defensiveness will be associated with measures of anxiety, as the literature cited earlier suggests. Which features of anxiety might be most closely associated with sensory defensiveness is an open question, as is the question of any association with features of temperament other than anxiety. Finally, little empirical evidence exists to guide hypothesizing about genetic and environmental underpinnings of tactile and auditory defensiveness. Variation in other traits involving reactivity and regulation of behavior in early development does show genetic effects (Goldsmith, 2003), and the physiological correlates of sensory defensiveness also render a genetic basis plausible. However, biology can be influenced by experience, and tactile defensiveness can be induced in nonhuman primates by exposure to certain agents (Schneider, 2004). Thus, there is reason to expect both genetic and environmental influences on our measures.

METHOD

Participants

The sample comprised families of young twins recruited for the Wisconsin Twin Project, a statewide, birth register-based panel (Van Hulle, Lemery, & Goldsmith, 2002). The names and contact information for all families were provided by the state for birth years 1998, 1999, and 2000. Families were invited to join the study shortly after the twins were born. The response rate was 76.5% for uncompensated participation. The sample of 1,394 twins comprised 225 monozygotic (MZ) and 458 dizygotic (DZ; 237 same sex) twin pairs, with 14 pairs of uncertain zygosity (see later).

Zygosity and temperament surveys as well as questions about medical history and demographics were administered during a telephone interview with the primary caregiver. The Infant-Toddler Social and Emotional Assessment (ITSEA; Carter & Briggs-Gowan, 2000) was sent to the families for completion by both parents. Approximately 65% of eligible families completed the telephone interview. Of those, 67% of mothers and 62% of fathers returned the mailed questionnaires, resulting in 756 twins for analyses involving ITSEA data.

The sample of twins was 47.9% females; the mean age at the time of assessment was 26 months, ranging from 11 to 36 months; 87.1% were between 24 and 30 months of age, with 8 pairs younger than 24 months and 79 pairs (mostly families who were harder to contact) older than 30 months. Thus, the study was not designed to examine variation in sensory defensiveness associated with age. The twins' race was reported as 90.9% Caucasian, 2.6% African American, 0.1% Native American, 0.4% Asian American, and 5.9% biracial. Mothers' educational attainment at the time of the twins' birth was as follows: 1.5% did not graduate from high school, 21.2% were high school graduates, 31.9% had one to three years of college, 30.7% were college graduates, and 14.5% had some graduate education.

Some analyses were restricted to a subsample of twin pairs in which at least one twin scored high on sensory defensiveness. The subsample ($n = 140$) contained 41 MZ and 99 DZ (43 same sex) twin pairs. The racial composition of the subsample was 85.4% Caucasian, 8.5% African American, and 6.2% biracial. Mothers' educational attainment was 30.8% high school graduates, 35.1% with one to three years of college, 20.8% college graduates, and 12.3% with some graduate education.

Measures

Zygosity

The Zygosity Questionnaire for Young Twins (Goldsmith, 1991) diagnosed zygosity and was administered by phone so that responses could be clarified. This questionnaire yields over 95% agreement with zygosity determined via genotyping (Forget-Dubois et al., 2003; Price et al., 2000). If parent responses did not result in a clear assignment of zygosity, a photo was requested, and, when possible, hospital pathology reports on the placenta(e) were obtained. The 14 pairs (2%) for whom zygosity could still not be unambiguously determined were excluded.

Sensory Defensiveness

Sensory defensiveness was assessed via the Sensory Defensiveness subscale of the revised Toddler Behavior Assessment Questionnaire (TBAQ; Goldsmith, 1996). The TBAQ is a caregiver-report temperament measure that assesses temperamental dimensions of 18- to 36-month olds. We used a revised 120-item version with the following subscales: Activity Level, Anger, Attention, Inhibitory Control, Interest, Object Fear, Pleasure, Sadness, Social Fear, and Soothability, plus Sensory Defensiveness. Par-

ents reported, on a 1 (*never*) to 7 (*always*) rating scale, how often they observed the specified behavior during the past month. A mean score (possible range 1–7) was calculated for each subscale. Alpha internal consistency reliability estimates ranged from .60 (anger) to .85 (inhibitory control).

The Sensory Defensiveness scale of the TBAQ comprises two 5-item subscales, Auditory Defensiveness and Tactile Defensiveness. The item content of the Sensory Defensiveness scale of the TBAQ (see Table 1) is similar to corresponding parts of the Sensory Profile (Dunn, 1999), a widely used instrument in sensory defensiveness research and clinical practice. The Auditory and Tactile Defensiveness subscales showed only moderate internal consistency ($\alpha = .57$ and $.51$, respectively), and the question of degree of independence of these two subscales is an issue treated in Results section.

Social–Emotional Behavior

Social–emotional behavior was assessed via the Infant–Toddler Social and Emotional Assessment (ITSEA; Carter & Briggs-Gowan, 2000; Carter, Briggs-Gowan, Jones, & Little, 2003). The ITSEA is a parent-report measure comprising 139 items that evaluate the following broad domains of social–emotional behavior of 1- to 3-year olds: Externalizing, Internalizing, Dysregulation, and Competencies. Internal consistency reliability estimates for the domains range from .86 to .90, and test–retest reliability estimates for the domains range from .82 to .90. The ITSEA also includes several narrower subscales, some of which were considered in this study. One of these narrower subscales is a three-item assay of Sensory Sensitivity. Many ITSEA items directly reflect symptoms in the *Diagnostic and Statistical Manual of Mental Disorders*, fourth edition, text revision (*DSM-IV-TR*, American Psychiatric Association, 2000).

Statistical Analyses

To characterize sensory defensiveness symptoms in this population, we conducted both phenotypic and genetic analyses. Correlational analyses assessed the association of sensory defensiveness symptoms with normal range temperament as well as problem behaviors. In these analyses, we treated the twins as individuals, without consideration of the dependency created by including cotwins in the same analyses, but we also conducted multilevel regression analyses that took this dependency into account. To clarify the clinical implications of sensory defensiveness, we examined the proportion of extremely sensory

Table I. TBAQ-R Sensory Defensiveness Item Content

Auditory items	
How often did your child seem to be alarmed when s/he heard sirens (such as a police, fire, or an ambulance siren) in the distance?	
How often did your child ask or gesture for the volume of loud music, radio, or TV to be lowered?	
How often did your child seem overly sensitive to, or irritated by, certain sounds, voices, or music?	
How often was your child distracted by background sounds that do not bother most other people?	
How often did your child react noticeably when a low-pitched sound started suddenly (such as an air conditioner, a heating system, a refrigerator, or a vacuum from another room)?	
Tactile items	
How often did your child object to scratchy clothing fabrics such as wool?	
When touching a new object, how often did your child seem concerned by how smooth or rough the texture was?	
How often did your child object to changes in articles of clothing that fit snugly or tightly (e.g., putting on a hat, wearing gloves, getting new shoes)?	
How often did your child refuse to touch a sticky or goopy substance (e.g., shaving cream, mayonnaise, toothpaste, mud)?	
How often did your child object to the feeling of a comb moving through her/his hair or a toothbrush touching her/his gums?	

defensive children who also scored in the top 10% on ITSEA behavior problem scales.

Comparing the resemblance between MZ and DZ cotwins allowed us to partition the individual differences on a trait into variation due to underlying genetic and shared and nonshared environmental factors, with a standard implementation of the twin method (Neale & Cardon, 1992). We used the program Mx (Neale, Boker, Xie, & Maes, 2001) for model fitting to the raw data. First, we parsed the variation in auditory and tactile defensiveness into underlying genetic and environmental factors. Then using a bivariate Cholesky decomposition, we analyzed the covariation between auditory and tactile defensiveness into common genetic and common environmental factors. These analyses permitted the estimation of univariate heritability (and shared and nonshared environmental influences) along with genetic and environmental covariation between the two traits.

Probandwise concordance rates were calculated for MZ and DZ twins separately. The concordance rate reflects the ratio of twin pairs with both twins affected to twin pairs in which at least one member is affected. Higher concordance rates in MZ than in DZ twins indicate that affected status is partly heritable. We also report tetrachoric correlations, which use extreme/nonextreme status to infer correlations between latent continuous dimensions, assumed to underlie extreme/nonextreme status. We fit biometric models to these tetrachoric correlations.

RESULTS

Lack of Difference in Auditory and Tactile Defensiveness by Gender and Zygosity in the Full Population

Mean auditory and tactile defensiveness scores for boys versus girls and for MZ versus DZ twins were

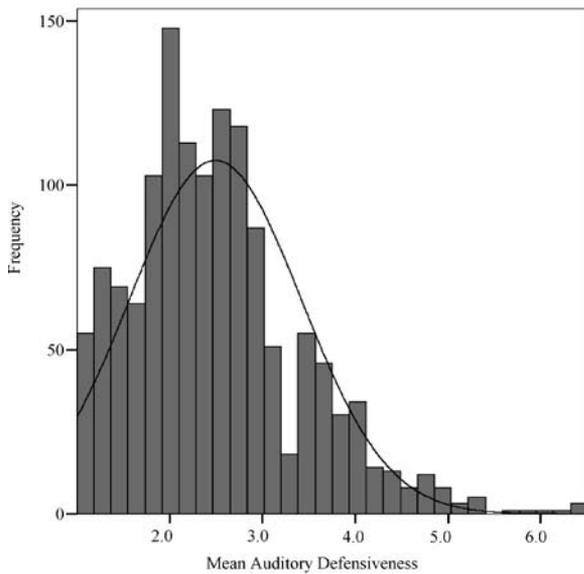
similar, and thus we pooled data across gender and zygosity. Given the large sample, we relied on the small group differences as a percentage of the standard deviation (the standardized effect size d ; Cohen, 1988) to justify this pooling. For auditory defensiveness, the gender mean difference was <5% of an SD (females: $M = 2.50$, $SD = 0.92$; males: $M = 2.46$, $SD = 0.90$; $p = .45$), and for tactile defensiveness, the gender mean difference was <9% of an SD (females: $M = 2.94$, $SD = 0.96$; males: $M = 2.86$, $SD = 0.95$; $p = .10$). For sensory defensiveness combined (mean of auditory and tactile), the gender mean difference was (females: $M = 2.71$, $SD = 0.77$; males: $M = 2.67$, $SD = 0.76$; $p = .24$).

For auditory defensiveness, the largest mean difference between zygosity groupings was < 14% of an SD (MZ twins: $M = 2.53$, $SD = 0.86$; same-sex DZ twins: $M = 2.41$, $SD = 0.94$; opposite-sex DZ twins: $M = 2.51$, $SD = 0.93$; $p = .09$). For tactile defensiveness, the largest mean difference between zygosity groupings was < 13% of an SD (MZ twins: $M = 2.93$, $SD = 0.93$; same-sex DZ: $M = 2.81$, $SD = 0.95$; opposite-sex DZ twins: $M = 2.95$, $SD = 0.98$; $p = .05$). For sensory defensiveness combined, the results were very similar (MZ twins: $M = 2.72$, $SD = 0.71$; same-sex DZ twins: $M = 2.60$, $SD = 0.76$; opposite-sex DZ twins: $M = 2.72$, $SD = 0.77$; $p = .02$).

Distribution of Auditory and Tactile Defensiveness

As shown in Panel A of Fig. 1, the scores for auditory defensiveness were relatively normally distributed in our population (Shapiro-Wilk's test = .96), with the exceptions of a lack of "no symptom" cases on the left side of the distribution and the occurrence of some cases rated as very defensive, or beyond the right-sided tail of the curve. As shown in Panel B of Fig. 1, a similar distribution was obtained for tactile defensiveness (Shapiro-Wilk's test = .99). Appropriate cutoff points for extreme scores were

PANEL A: Auditory Defensiveness



PANEL B: Tactile Defensiveness

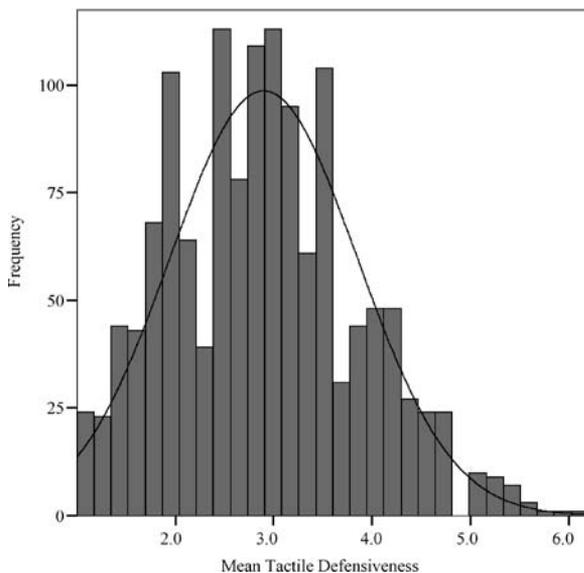


Fig. 1. Distribution of sensory defensiveness scores.

not readily apparent from these distributions. However, a pilot study by Ahn, Miller, Milberger, and McIntosh (2004) indicated that approximately 5% of children might be affected by sensory defensiveness impairment. The 5% threshold yields a reasonable number of extreme cases for analyses, and it also captures all individuals whose scores might be considered as lying beyond the tail of the curves. Therefore, the top 5% of individuals in the auditory ($N = 67$) and tactile ($N = 71$) and combined ($N = 76$) de-

fensiveness distributions were considered to be “high” on sensory defensiveness for purposes of our analyses.

A cross-tabulation analysis indicated that only 7 children (out of 67 for auditory defensiveness and 71 for tactile defensiveness) were in the extreme group on both dimensions. Although this degree of association shows a trend toward statistical significance (chi square with continuity correction = 2.853, $df = 1$, $p = .091$), the two extreme groups had only 9.9–10.4% overlap, depending on which N is taken as the denominator. This finding is consistent with the moderately low correlation between the two subscales reported later. The gender distribution for the high tactile defensive group was 44 (62.5%) females and 27 males; for the high auditory defensive group, it was 32 (47.1%) females and 37 males. A two-tailed binomial test revealed that a significant majority of those who displayed extreme tactile defensiveness were girls ($p = .058$).

Excluding the possibility that sensory defensiveness was secondary to medical issues was important to the interpretation of our findings. Primary caregivers responded to an open-ended question asking about the occurrence of serious illnesses, accidents, and medical complications for each twin during the phone interview. Of all individuals who scored above the 5th percentile for either type of sensory defensiveness, 10% of the auditory and 14% of the tactile group, reported medical problems, serious illnesses, or accidents. The types of complications reported were not different from those for children who were below the 5th percentile, nor were there apparent connections between the complications reported and auditory and tactile sensitivities. For example, more chronic ear infections were reported in the high tactile defensiveness group than in the high auditory defensiveness group. Of those twins who were either tactile or auditory defensive, 62% were born prematurely (less than or equal to 36 weeks), whereas 56% of those without tactile or auditory defensiveness were born prematurely. The difference in weeks gestation for the defensive and nondefensive groups was not significant, $F(1, 1,344) = 1.997$, $p = .16$, although this difference would qualify as a trend ($p < .10$) with a one-tailed test. Confirming this very modest degree of association with prematurity, sensory defensiveness scores in the entire sample were correlated $-.07$ with weeks of gestation (a posteriori $p = .01$, $N = 1,349$), with this very small effect largely accounted for by the auditory subscale.

Sensory defensiveness is a common feature of autism (Baranek, 1999a), and our findings would have a different interpretation if most children above the 5th percentile qualified for an autism spectrum diagnosis. Given the young age of our sample, definitive statements about diagnosis on the autism spectrum are impossible. However,

we employed three autism screening items that were part of the ITSEA, “Points to show you something far away” (reverse scored for autism risk), “Pretends that objects are something else. For example, uses banana as phone” (reverse scored for autism risk), and “Does not make eye contact.” These items were each scored on a 0–2 scale, yielding a possible range of 0–6 for the autism risk indicator. On the basis of data from Carter and Briggs-Gowan (2000), we regarded a score of 4 or above as indicating a notable risk for autism. The autism screening items of the ITSEA were completed by mail, and are not available for the entire 5% of the auditory and tactile distributions. For the auditory defensive group ($n = 36$), only five (14%) individuals scored 4 or above on the autism items. For the tactile defensive group ($n = 48$), only five (10%) of the individuals scored 4 or above on the autism items. Although rates of 14 and 10% are above the population risk for autism screening items (Carter & Briggs-Gowan, 2000), as would be expected for any component phenotype of autism, it is clear that the great majority of children above the sensory defensiveness thresholds do not screen positive for autism, nor do the extreme children show the high male:female ratio characteristic of autism.

Association Between Auditory and Tactile Defensiveness

The correlation of the Auditory and Tactile Defensiveness subscales of the TBAQ was moderate, $r(1,357) = .29$, $p = .01$, suggesting that each might carry substantial independent information. However, the modest reliabilities of the measures make conclusions about independence from the phenotypic correlation tentative. On

the basis of the modest interscale correlation, the minimal overlap of extreme group membership, and the differences in gender ratios, we considered it prudent to retain the separate auditory and tactile scores for further data analyses. However, because these data hardly compel a conclusion that the two domains are distinct, we also considered it prudent to form an overall sensory defensiveness score (mean of the two 5-item subscales) for further analyses.

Association of Sensory Defensiveness with Temperament (TBAQ)

Analyses of the Full Sample

Table II displays the phenotypic correlations between the combined sensory defensiveness scale and auditory and tactile subscales and the other TBAQ scales. As anticipated by the literature, the largest correlations involved fear-related aspects of temperament. In particular, Object Fear was correlated the three sensory defensiveness scales. Importantly, 3 of the 10 Object Fear items reference behaviors that may be related to auditory processing difficulties (i.e., “During a loud storm, how often did s/he look afraid?”). These items would artificially inflate correlations with the auditory defensiveness scale and to a lesser degree with the overall sensory defensiveness scale, but not with the tactile defensiveness scale. Children high on sensory defensiveness were slightly more likely to be reported as expressing more social fear (shyness), sadness, and anger, as well as being more active and less soothable. Table II offers little evidence for differential correlations of tactile and auditory defensiveness with temperamental dimensions.

Table II. Correlations of Sensory Defensiveness Scale and Subscales with TBAQ Scales

TBAQ scales	Overall sensory defensiveness	Auditory defensiveness	Tactile defensiveness
Attention	-.09**	-.08*	-.06
Interest	-.03	-.02	-.02
Activity level	.15***	.10***	.16***
Inhibitory control	-.12***	-.08**	-.12***
Sadness	.25***	.17***	.24***
Anger	.22***	.15***	.19***
Object fear	.50***	.42***	.36***
Social fear	.19***	.13***	.18***
Pleasure	-.02	.04	-.08**
Soothability	-.24***	-.18***	-.20***

Note. $N = 1,075$.

* $p < .05$. ** $p < .01$. *** $p < .001$.

Association of Sensory Defensiveness with Behavior Problems (ITSEA)

Preliminary Analysis: Formation of Mid-Parent ITSEA Scale Scores

Among the subsample in which at least one parent returned the mailed ITSEA, both parents responded in 75.7% of cases. Correlations between mother and father report ranged from .26 to .56, with a mean of .42. Given this of level of parental agreement, a mid-parent mean was computed by averaging the parents' scores. Differences between the mid-parent report and maternal report, calculated on the same cases, ranged from 3 to 16% of a standard deviation. Thus, the mid-parent mean was used in all analysis involving ITSEA data, but in the cases where paternal report was unavailable, maternal report only was used.

Analyses Based on Dimensional Scores

Only 378 of the 694 families who provided information by telephone about their children's sensory defensiveness returned a subsequent questionnaire packet containing the ITSEA. Sensory defensiveness scores in this subsample were very similar in mean and variance from the group who did not return the questionnaire packet. Even with the large sample size, no variance differences approached the criterion for statistical significance. The means for sensory defensiveness were slightly higher in the group whose parents did not return the questionnaire packet (18, 1, and 14% of a standard deviation for auditory, tactile, and combined sensory defensiveness, respectively).

The data in Table III are presented as correlations to ease interpretation of the results. However, these analyses ignore the dependence in twin data; that is, cotwins in a pair are treated as though they were independent observations. This might not be a serious violation of assumptions but it does suggest the need for further investigation. Various regression-based approaches using generalized estimating equations allow adjustment for dependence of observations in circumstances such as these (Hardin & Hilbe, 2003). (Our dependence is due to twin pair membership; in other circumstances, the dependence might arise from repeated measures or from other clustering such as students clustered in different schools.) We thus used SAS PROC GEN to reexamine the correlations in Table III using multilevel regression. We examined the parameter estimates (regression coefficients) generated by the generalized estimating equations for the association of tactile or auditory defensiveness and their combination with the

ITSEA symptom scales as well as the empirical standard errors of each estimate.

Overall Internalizing symptoms (fourth row in Table III) continued to be significantly associated with all three sensory defensiveness scales (tactile, auditory, and combined), $ps < .001$. Eight of the nine possible associations among the internalizing subscales (Depression/Withdrawal, General Anxiety, and Separation Distress) and the three sensory defensiveness scales were significant in the uncorrected analyses (rows 5 through 7 in Table III). With the multilevel regression approach, five of these eight associations continued to be significant, $ps < .05$, and the other three were reduced to trends (ps ranging from .05 to .10).

In the multilevel regression analyses, overall Externalizing symptoms (third row in Table III) continued to be significantly associated with the tactile and combined sensory defensiveness scales, $ps < .05$; however, the association with the auditory defensiveness scale was nonsignificant, $p = .21$. Examination of Table III shows 15 significant correlations of Dysregulation, Negative emotionality, Eating, Maladaptive symptoms, Repetitive movements, and Social relatedness with either overall sensory defensiveness or the auditory or tactile domains. After correction for dependence in the multilevel regression analyses, 13 of these 15 associations remained significant ($p < .05$); one was reduced to a trend ($p = .06$); and one was not significant. The exception to this pattern of replication was the Sleep symptom ITSEA scale, where the uncorrected correlations (rs ranging from .08 to .10) could not be confirmed as significant associations after the correction for dependence. Thus, we conclude from these subsidiary analyses that the dependency in the twin data did not substantially bias the results. Full results of the SAS PROC GEN analyses are available from the authors.

Association of Sensory Defensiveness with Clinically Significant Levels of Impairments

Correlations in the full sample do not necessarily address the issue of whether the children most impaired by behavioral problems are also extreme in sensory defensiveness. Without clinical diagnoses, we can only offer an initial approach to this issue. Although empirically validated cutting scores to predict clinical status from ITSEA are still under development (A. Carter, personal communication, March 17, 2004), scores in the upper 10% of the distribution designate reasonable risk groups. Thus, we calculated the percentage of children with high (top 5%) sensory defensiveness scores who also scored in the top 10% on the major ITSEA behavior problem scales (see Table III). A one-tailed, z -approximation test

Table III. Correlations of Sensory Defensiveness Scale and Subscales with Selected ITSEA Scales

	Overall sensory defensiveness		Auditory defensiveness		Tactile defensiveness	
	Zero-order correlation	% ^a	Zero-order correlation	% ^b	Zero-order correlation	% ^c
Externalizing symptoms	0.14***	20.6	0.10**	17.2	0.12***	19.4
Internalizing symptoms	0.22***	28.1**	0.16***	22.6*	0.19***	22.5**
Depression/Withdrawal	0.15***	21.8*	0.11**	16.1	0.13***	20.0*
General anxiety	0.28***	31.2***	0.24***	25.8**	0.21***	20.0*
Separation distress	0.10**	21.8*	0.11***	16.1	0.06	15.0
Dysregulation	0.29***	31.2***	0.18***	35.5***	0.28***	10.0
Sleep	0.10**	28.1**	0.08*	22.6*	0.08*	15.0
Negative emotionality	0.15***	37.5***	0.09*	25.8**	0.15***	27.5***
Eating	0.10**	21.8*	0.02	19.4	0.14***	30.0***
Maladaptive	0.18***	31.2***	0.23***	38.7***	0.05	7.5
Repetitive movements	0.19***	18.8	0.17***	12.9	0.13***	12.5
Social relatedness	-0.08*	12.5	-0.05	9.7	-0.08*	10.0
Competence	-0.03	10.0	0.01	10.0	-0.07	0.0
Atypical index	0.04	9.4	0.06	0.0	0.00	15.0

Note. $N = 1,075$. The z -approximation tests were used to determine whether observed proportions of high (top 5%) sensory defensiveness children scoring within the top 10% on ITSEA scales differed significantly from hypothesized population proportion of .10. See text for discussion of related multilevel regression analyses.

^aThe percentage indicates proportion of children high on sensory defensiveness who score in the top 10% on ITSEA scales.

^bThe percentage indicates proportion of children high on auditory defensiveness who score in the top 10% on ITSEA scales.

^cThe percentage indicates proportion of children high on tactile defensiveness who score in the top 10% on ITSEA scales.

* $p < .05$. ** $p < .01$. *** $p < .001$.

assessed whether the estimated proportion differed significantly from 10%. Results suggested that children high on combined sensory defensiveness may be at increased risk for developing internalizing problems, dysregulation problems, and maladaptive problems. There is only minimal evidence of differential association of auditory and tactile defensiveness with behavior problems. On the other hand, highly sensory defensive children were not significantly overrepresented among toddlers high on externalizing symptoms, social relatedness, competence, and the atypicality index. Even for the internalizing symptoms, about 70% or more of highly sensory defensiveness children did not score in the high symptom range.

Genetic and Environmental Influences on Auditory and Tactile Defensiveness and Their Overlap

Univariate Analyses

Because of the lack of mean gender differences or significant differences in variances across zygosity (see

analyses earlier), MZ and DZ groups were pooled across genders for the correlational analyses. The second and third columns of Table IV show the correlations indexing MZ and DZ twin similarity for auditory and tactile defensiveness. In both cases, MZ twins were more similar than were DZ twins, leading us to infer some genetic influence. This MZ – DZ difference was greater for tactile defensiveness, suggesting stronger genetic influence.

Structural equation modeling was used to quantify these impressions from the twin correlations. To maximize power, models were fit directly to raw data. For auditory defensiveness, genetic and shared environmental factors were significant. Heritability was calculated at 38%, shared environment accounted for 33% of the variance, and nonshared environmental factors accounted for 28%. For tactile defensiveness, the heritability was 52%, and nonshared environment accounted for 31% of the individual variation. The shared environment parameter, while accounting for 17% of the individual variation, only barely reached a conventional significance criterion (dropping this parameter resulted in a chi-square difference

Table IV. Twin Similarity Correlations and Biometric Model Fitting Results for Auditory and Tactile Defensiveness

Sensory defensiveness domain	r_{MZ}	r_{DZ}	h^2	c^2	e^2
Auditory	.72	.51	.38 (0.22, 0.54)	.33 (0.19, 0.48)	.28 (0.23, 0.34)
Tactile	.69	.40	.52 (0.34, 0.71)	.17 (0.01, 0.33)	.31 (0.26, 0.37)

Note. r_{MZ} : MZ intraclass correlation; r_{DZ} : DZ intraclass correlation; h^2 : heritability; c^2 : variance accounted for by shared environment; e^2 : variance accounted for by nonshared environment; 95% confidence intervals are given in parentheses.

of 4.0, $p = .04$). Because twins are of the same age, and often the same gender, any age and gender effects are likely to contribute to the shared environment variance. Univariate models were refit to the data after residualizing on age, gender, and their interaction; however, there were no notable changes in the resulting parameter estimates.

Bivariate Analyses

We next analyzed the phenotypic association of auditory and tactile defensiveness from a biometric perspective. The intraclass cross-twin, cross-domain correlations were similar for MZ and DZ twins ($r_{MZ} = .28$, $r_{DZ} = .25$), suggesting only modest genetic influence on the cross-domain association. A bivariate Cholesky model was used to test for common factors underlying both auditory and tactile defensiveness. The bivariate model provides a more powerful test of genetic and environmental influences than does univariate model (Schmitz, Cherny, & Fulker, 1998). To test the significance of pathways common to both traits, the fit of the full model was compared with submodels that dropped the pathway of interest. The significant pathways and their estimates are shown in Fig. 2. The genetic factors that are common to both auditory and tactile defensiveness were not significant. Thus, the majority of individual variation in auditory and tactile defensiveness was due to specific genetic factors. In contrast, a single shared environmental factor influenced both tactile and auditory defensiveness. Interestingly, shared environment influences on tactile defensiveness are higher in the bivariate model than the univariate model (22% vs. 17%). As might have been expected, nonshared environmental effects were unique to each domain.

Genetic Influences on Extreme Auditory and Tactile Defensiveness

To begin examining genetic effects for individuals who might have clinically significant elevations on sen-

sory defensiveness, probandwise concordance rates were calculated for MZ and DZ twins (Table V). The upper third of Table V presents data for the extreme samples, containing only the top 5% on the Auditory Defensiveness or Tactile Defensiveness scale. For both auditory and tactile defensiveness, the concordance rates were higher among MZ than DZ twins. This MZ–DZ discrepancy was greater for tactile defensiveness than for auditory defensiveness, suggesting that heritable influences might be stronger for extreme levels of tactile defensiveness. Examination of the data revealed that many cotwins missed the 5% cutoff by a small margin. Therefore, the probandwise concordance rates were also calculated using scores from the probands in the upper 5% of the distribution, but with the cutoff for the cotwin's score relaxed to the top 15% of the distribution (see middle third of Table V). MZ concordance rates increased more than did DZ concordance rates. This suggests not only stronger genetic effects for both traits but also greater differential heritability (extreme tactile defensiveness more heritable than is extreme auditory defensiveness). Another way to view this issue is by calculating tetrachoric correlations for extreme status; these correlations are presented in the lower third of Table V.

Heritability of the extreme group can be estimated using a liability threshold model, which assumes that the trait has an underlying normal distribution but is only identifiable in persons who surpass some threshold. As before, we assumed the presence of a single threshold that identified cases in the top 5% of the distribution for both auditory and tactile defensiveness. Models were fit to the raw proband status (above threshold vs. below threshold). For auditory defensiveness, familial influences were significant ($h^2 = .42$, $c^2 = .30$, $e^2 = .27$) though it was not possible to distinguish between a model that included only heritable factors ($\Delta\chi^2 = 1.0$, $\Delta df = 1$, $p = .31$) and a model that included only share environmental factors ($\Delta\chi^2 = 1.1$, $\Delta df = 1$, $p = .30$). In contrast, for tactile defensiveness only heritable and nonshared environmental influences were significant ($h^2 = .76$, $c^2 = 0$, $e^2 = .23$).

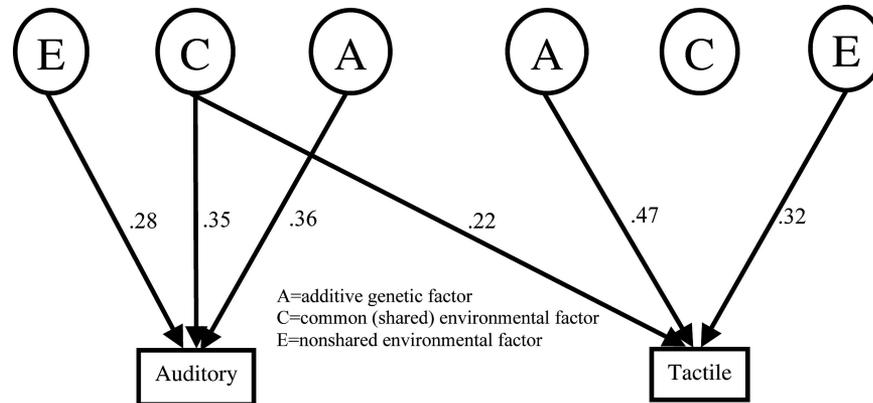


Fig. 2. Bivariate Cholesky model for the association between auditory and tactile defensiveness, with genetic and environmental parameter estimates

DISCUSSION

In this initial twin study of sensory defensiveness, we addressed questions about the distribution of sensory defensiveness in our population-based sample, questions about association with other behavioral problems and traits, and quantitative genetics questions. In brief, scores were continuously distributed with peaks in the moderate range and with some accumulation of high scores—especially for auditory defensiveness—but no clear bimodality that would unambiguously suggest subgroups of “affected” and “unaffected” individuals. Our hypothesis that both auditory and tactile defensiveness would be correlated with fearful temperament and anxiety was supported. Differences in tactile and auditory defensiveness showed moderate genetic influences, with tactile defensiveness demonstrating somewhat greater heritability in each of the several ways of evaluating heritability. Perhaps the major contribution of the study is a simple one: it is the first study to demonstrate familial aggregation of sensory defensiveness.

Limitations

Generalization and interpretation of our results are limited by certain design factors. First, all information was obtained via parental report. Although standard in the field, parental report questionnaires have certain limitations (Rothbart & Goldsmith, 1985). Nevertheless, parental report can be defended as economical, thus allowing very large samples such as ours, and particularly reasonable for studying sensory defensiveness. Parents observe their children in a wide range of situations and are likely to witness rare behaviors that otherwise could not be assessed. Given that a noxious material or sound might subsequently be avoided, some sensory defensive behaviors could be infrequent. Also, parents are the likely recipients of complaints about sensory issues in toddlers. Finally, we suggest that sensory defensive behaviors are not markedly low in social desirability; parents do not appear to be embarrassed or reticent to report sensory defensiveness. Thus, we do not regard the limitation of assessment of sensory defensiveness to parent report as

Table V. Probandwise Twin Concordance Rates and Tetrachoric Correlations for Domains of Sensory Defensiveness

Sensory defensiveness domain	Zygosity	
	MZ	DZ
Concordances: 5% criterion for probands and cotwins		
Auditory defensiveness	6/17 (35%)	14/52 (27%)
Tactile defensiveness	12/24 (50%)	6/47 (13%)
Concordances: Relaxed criterion for cotwins only		
Auditory defensiveness	11/17 (65%)	26/52 (50%)
Tactile defensiveness	20/24 (83%)	15/47 (32%)
Tetrachoric correlations: 5% criterion used to dichotomize the distribution		
Auditory defensiveness	.72 (0.32–0.92 CI)	.53 (0.26–0.73 CI)
Tactile defensiveness	.82 (0.56–0.95 CI)	.27 (–0.07–0.56 CI)

problematic although we acknowledge that the prior literature validating parental report with behavioral measures of sensory defensiveness is sparse. However, several studies in the temperament domain have shown that parents can be reliable reporters of their children's behavior (Bishop, Spence, & McDonald, 2003; Goldsmith & Rothbart, 1991; Matheny, 1987). Although some of our parent measures came from telephone interviews and some from questionnaires, it is also true that correlations with other domains (e.g., anxiety) could be inflated by common method variance. We dealt with this possible contamination by using both maternal and paternal report on some measures (see Method section).

A second limitation is the modest internal consistency of our two 5-item subscales of sensory defensiveness. In the design phase of our study, it was not yet apparent that the auditory and tactile domains would need to be measured separately, and thus we did not anticipate this issue. Of course, 5-item scales reflecting complex behavioral domains cannot be expected to be highly internally consistent; therefore, we must ask how modest reliability might affect interpretation of results. Essentially, demonstrations of significant links with other domains and of heritability are realized despite limited internal consistency of the measures, and thus we can be confident in these demonstrations. However, interpretations of independence, or lack of associations, must be more guarded because modest internal consistency can limit estimates of associations with other measures.

The children in this study were toddlers, averaging 26 months of age. Because the developmental course of sensory defensiveness has not been characterized well empirically, generalization to other ages is unjustified. Yet another feature of the study is that the extreme individuals do not have a clinical diagnosis. This feature is not so much a limitation as a typical difference between clinic-based and community-based samples, which often provide different perspectives on behavioral problems. A final caution about our design is that we do not know if twins and singletons differ in sensory defensiveness although there is no obvious reason to expect such differences.

Generality Versus Domain Specificity of Sensory Defensiveness

The question of the generality versus domain specificity of sensory defensiveness might not have a simple answer. We first note that we studied only two domains of sensory defensiveness, and more specificity might be apparent in other domains such as vision, olfaction, taste,

vestibular function, and proprioception. The strongest evidence favoring commonality between auditory and tactile defensiveness was the similarity of their external correlates in the temperament and behavior problem domains. The evidence regarding independence of our measures of auditory and tactile defensiveness was somewhat ambiguous. Scales measuring auditory and tactile defensiveness were modestly but significantly correlated, and the overlap of extreme group membership was significant but included only a minority (about 10%) of the extreme cases. Girls more frequently showed tactile defensiveness, but not auditory defensiveness.

The strongest evidence for domain specificity emerged from the genetic analyses. Bivariate genetic analysis suggested different genetic factors for individual differences in auditory and tactile defensiveness. In addition, tactile defensiveness was somewhat more heritable for both the full range of scores and for the extreme groups. One reason for this difference might be that the auditory environment tends to be imposed on the child, indeed on both twins, whereas the tactile environment can be chosen or avoided, to some degree, by an individual (i.e., children can choose to touch some objects or materials and avoid others or reject some clothing items and accept others). Therefore, we might expect a greater role of the individual and greater heritability for tactile defensiveness than for auditory defensiveness. In other words, tactile defensiveness may be subject to active gene-environment correlation to a greater degree than is auditory defensiveness.

Implications for Prenatal Effects

Shared environmental factors were modest but significant contributors to similarity for auditory and tactile defensiveness. Twins, regardless of zygosity, share the same prenatal environments. Therefore, if exposure to prenatal stressors is a "main effect" (i.e., if the stressors do not interact with genetic susceptibility factors), then we would expect that shared environmental factors would emerge as significant contributors to individual differences. The shared environmental factor was only marginally significant in the present sample for tactile defensiveness; thus, our results do not strongly implicate the prenatal environment in tactile defensiveness in the absence of genetic susceptibility factors. This conclusion is qualified by power limitations in demonstrating shared environmental factors with our sample size. Moreover, these twin-based results do not contradict the finding that prenatal lead exposure can result in tactile defensiveness in rhesus monkeys (Schneider, 2004), because lead exposure is presumably rare in the pregnancies of twins in

our sample and thus could not affect the environmental variance components we estimated.

Implications for Wider Recognition of Sensory Modulation Challenges

As noted in Introduction section, sensory problems are a neglected domain in the *DSM-IV-TR* (American Psychiatric Association, 2000), without status as a diagnostic entity and not mentioned as a key symptom. However, the sensory modulation domain is included in the most recent version of the *Diagnostic Manual for Infancy and Early Childhood* (Interdisciplinary Council on Developmental and Learning Disorders Work Groups, 2005). This study offers modest evidence for sensory defensiveness, or over-responsiveness, as a relatively independent dimension or category. However, the children in this sample were too young to be diagnosed with, for example, ADHD. Thus, the independence of sensory defensiveness from other diagnoses cannot be firmly established by our data, despite the lack of substantial overlap with extreme behavior problem scale scores (see Table III). At the least, our results suggest that studying sensory modulation more systematically is justified.

Future Directions

These results suggest several needed studies. First, the genetic influences inferred from twin similarity should be confirmed in family studies, where evidence of vertical transmission could be gathered. Second, studies should include some in-person, objective measurement of reactions to exposure to noxious sounds and textures. Third, some genetically informative studies should be longitudinal in design, to allow examination of patterns of change and stability in sensory defensiveness. Fourth, studies should include sufficient samples of both persons and variables to allow taxometric analyses to determine whether sensory defensiveness is better conceptualized as a trait or a type. Additional population-based studies, such as ours, are also necessary to clarify how frequently significant levels of impairment occur independently and co-occur with other medical or behavioral challenges.

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