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Abstract

Background

Studies have shown that children with pervasive developmental disorder (PDD) have high rates of sensory hypersensitivity. In addition, a few recent studies suggested that sensory hypersensitivity was related to anxiety or depression. However, most studies had methodological limitations because they included children with mental retardation and did not examine broadband psychopathology. Therefore, the purpose of this study was to examine the prevalence of sensory hypersensitivity in children with high-functioning PDD (HFPDD) and the correlation among sensory hypersensitivity, various characteristics, and broadband psychopathology.

Methods

We assessed 132 children with HFPDD (aged 6-15 years, 75% male) that were divided into sensory hypersensitivity (HS) and sensory non-hypersensitivity (non-HS) groups. A logistic regression model was used to examine correlations among sensory hypersensitivity, age, gender, PDD subtypes, socioeconomic status, and broadband psychopathology, including symptoms of anxiety and depression.

Results

Of the 132 children with HFPDD, 65.9% (n=87) were categorized as HS and 34.1% (n=45) as non-HS. The most common sensory hypersensitivity was auditory. Logistic regression analyses revealed that sensory hypersensitivity in HFPDD was significantly associated with autistic disorder and symptoms of anxiety and depression.

Conclusions

Majority of children with HFPDD exhibited sensory hypersensitivity. Our findings suggested that sensory hypersensitivity may be a core feature of HFPDD and is possibly correlated to symptoms of anxiety and depression. We propose that sensory hypersensitivity in children with PDD should be aggressively assessed.

Key Words: Pervasive developmental disorder; Sensory hypersensitivity; Children; HFPDD; CBCL

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Introduction

Pervasive developmental disorder (PDD) is characterized by qualitative impairments in social interactions and communication skills, along with a restricted repetitive and stereotyped behavior pattern that are most frequently diagnosed on the basis of behavioral criteria described in the Diagnostic and Statistical Manual of Mental Disorders, 4th Edition, Text Revision (DSM-Ⅳ-TR) of the American Psychiatric Association[1]. PDD refers to a group of three disorders, including autistic disorder, Asperger's disorder, and pervasive developmental disorder - not otherwise specified (PDD-NOS). PDD are recognized as a continuum or spectrum of these disorders, from most (autistic disorder) to least (PDD-NOS) severe and are marked by various intellectual levels, in which PDD without mental retardation is known as high-functioning PDD (HFPDD).

In addition to the core impairments of PDD, many researchers have reported that afflicted children frequently display sensory abnormalities[2-16] that occur across all sensory domains (auditory, visual, tactile, gustatory, and olfactory) and include hypersensitivity and hyposensitivity to sensory stimuli. For example, sensory hypersensitivity may include intolerance of auditory stimuli, such as appliance noise, loud gymnasiums, or various humming or crackling sounds that most subjects find inoffensive, and sensory hypersensitivity in the tactile domain that may occur in response to particular stimuli, such as certain fabrics, clothing tags, or viscous food textures. Leekam et al[7] reported that children with PDD displayed sensory abnormalities and symptoms with regard to multiple sensory domains.

Abnormal sensory responses among children with PDD can affect participation in daily activities, academic achievements, and social interactions[17-19], and sensory challenges may also negatively impact the early development of important relationships and positive social participation with schoolmates. Furthermore, aversive sensory experiences may impact the ability to master a range of essential developmental tasks and lead to impaired functioning[20]. For example, children with sensory hypersensitivity may find typical childhood interactions too loud; therefore, they may be unable to engage in age-appropriate interactions in a school lunchroom or on the playground.

Many children with PDD reported comorbid disorders[21-25], including anxiety and depression, which may further contribute to the avoidance of social situations and awkward interactions with peers, thus promoting further isolation from peers of the same age. Furthermore, recent findings have revealed a link between sensory abnormalities and anxiety and depression[6,17,26,27].

Overall, previous results suggested that sensory hypersensitivity was a common symptom in children with PDD and may be associated with other psychopathologies such as anxiety and/or depression. However, these studies had methodological limitations. First, they included children with mental retardation [intelligence quotient (IQ) <70], who generally have more psychiatric symptoms than those without mental retardation[28]. We reasoned that if children with PDD and mental retardation are included in a study, it may result in a higher prevalence of psychopathologies than studies including only children with HFPDD (IQ $\geqq 70$). Secondly, some studies were conducted without considering the severity of PDD. However, no previous studies have examined the association between sensory hypersensitivity and clinical characteristics, including PDD subtypes and broadband psychopathologies, in children with HFPDD. Therefore, in the present study, we assessed the association among sensory hypersensitivity, PDD subtypes, age, gender, IQ, socioeconomic status (absence of a parent; receipt of public assistance), and broadband psychopathology, including anxiety and depression, in children with HFPDD.

Methods

Subjects

The study subjects included 132 elementary or junior high school students with HFPDD, aged 6-15 years, who were consecutively referred to the children's psychiatry outpatient clinic of Osaka City University Hospital (Osaka, Japan) between January 2003 and August 2011.

PDD diagnoses were confirmed by experienced child psychiatrists and based on DSM-Ⅳ-TR criteria, a comprehensive developmental history, a clinician's interview with the child and the child's caregivers, and direct observations of the child. PDD-NOS was defined as a residual category of PDD and had no operational criteria in the DSM-Ⅳ-TR criteria[1]. Thus, we used the Buitelaar and van der Gaag diagnostic criteria for PDD-NOS, which required a total of three or more items from criterion A of the DSM-Ⅳ-TR criteria for autistic disorders, including at least one item identifying a qualitative impairment in social interactions[29]. In the present study, we excluded children with mental retardation (IQ <70; WISC-Ⅲ, Wechsler Intelligence Scale for Children-Third Edition[30]) to assess subjective psychophysical impairments. Children with severe neurological impairments or intractable epilepsy were also excluded.

We explained the investigational purpose, procedures, potential risks, and alternatives to participation and obtained written informed consent from the subjects and/or their parents. The study protocol was reviewed and approved by the Human Subject Review Committee of Osaka City University.

Sensory hypersensitivity

Sensory hypersensitivity has been evaluated by various methods[2-16]; however, neither a universally accepted criteria nor a rating scale for sensory hypersensitivity have been established. Therefore, we defined sensory hypersensitivity as discomfort or irritability in response to non-noxious stimulation in a typical individual. All children and their parent(s) were independently interviewed to obtain information regarding the child's hypersensitivity in auditory, visual, tactile, olfactory, and gustatory sensory perceptions using semi-structured interview by child psychiatrists. For example, they were interviewed about such as exaggerated startle response to daily sounds, fear of minor smell and avoidance of certain fabrics. Teachers of the children were asked to complete a questionnaire or to participate in the telephone interview regarding the child's in-school sensory hypersensitivity by child psychiatrists. We obtained the information from 102 teachers (91% response). Based on all information on interviews, medical records and questionnaires, our assessment team involving a child psychiatrist blind to PDD subtypes classified sensory hypersensitivity (HS) as hypersensitivity in one or more domains at the time of consultation and sensory non-hypersensitivity (non-HS) as no hypersensitivity. Of the 132 children with HFPDD, 87 (65.9%) were assigned to the HS group and 45 (34.1%) to the non-HS group. Psychopathologies of the HS group were compared with those of the non-HS group.

Broadband psychopathology

We used the Child Behavior Checklist (CBCL) to assess the association between sensory hypersensitivity and broadband psychopathology, including symptoms of anxiety and depression (Itani et al[31] standardized a Japanese version of the CBCL, as developed by Achenbach et al[32]), which comprises 113 items across the following three domains: Internalizing, Externalizing, and Total Problems. Each item is rated on a three-point scale ranging from "not true" to "very true". The CBCL provides T scores and percentiles for three competence scales (Activities, Social, and School),

Total Competence, and Internalizing, Externalizing, and Total Problems. The CBCL provides eight empirically derived Syndrome Scales. The CBCL for subjects aged 6-18 years demonstrated good test/retest reliability, cross-informant agreement, and successful discrimination between referred and non-referred children[32]. The CBCL reportedly has strong psychometric properties as shown by samples of children with PDD[33].

Statistical analyses

All statistical analyses were performed using SPSS ver. 20.0 statistical software (SPSS Japan, Inc., Tokyo, Japan). The chi-squared test was used for categorical comparisons of the data. For continuous variables, normal distribution of the scores was examined. When the data were normally distributed, a *t*-test was used to examine intergroup differences. The Mann-Whitney *U*-test was used to assess data not normally distributed. A probability (p) value of <0.05 was considered statistically significant, and all statistical tests were two-tailed.

Logistic regression models were used to identify possible statistical associations among sensory hypersensitivity, age, gender, PDD subtypes, socioeconomic status (absence of a parent; receipt of public assistance), and broadband psychopathology in children with HFPDD. The odds ratio (OR) and 95% confidence intervals (95% CIs) were calculated after simultaneously controlling for potential confounders. Model variables included age, gender, IQ, PDD subtype, socioeconomic status, T scores of Internalizing, Externalizing, Total problems, and all eight Syndrome Scale scores on the CBCL. PDD subtypes were classified into two groups: autistic disorder or two related, but less, severe disorders: Asperger's disorder and PDD-NOS.

Initially, we conducted univariate analyses to examine potential relationships between independent and dependent variables using *t*-tests and chi-squared analyses for continuous and categorical independent variables, respectively. With regard to the selection of independent variables, we selected one or more variables from any measurement indicating a significant association ($p<0.05$) with dependent variable. Secondly, to assess multicollinearity, we calculated the Spearman's rank correlation coefficient between all variables. Since the T scores of Total, Internalizing, Externalizing on the CBCL showed strong correlations with the T scores of Syndrome Scales ($|r|>0.7$), we did not enter the T scores of Internalizing, Externalizing, and Total Problems into the logistic regression analyses. Finally, five variables (age, gender, PDD subtypes, and T scores of "Withdrawn" and "Anxious/Depressed" on the CBCL) were included into logistic regression analyses as independent variables.

Results

Of the 132 children (101 males and 31 females; mean age, 10.7 years; mean WISC-Ⅲ full-scale IQ score, 96.0) diagnosed with HFPDD, 97 were definitively diagnosed with autistic disorder, 33 with PDD-NOS, and two with Asperger's syndrome. Characteristics of all subjects and the HS and non-HS groups are shown in Table 1. Of the 132 children with HFPDD, 65.9% exhibited sensory hypersensitivity in at least one domain, and of these, 41.7% involved only one domain, 15.9% involved two, 7.6 % involved three, 0.8% involved four, and none showed any relation to all the five domains (Table 2). The most common sensory hypersensitivity was auditory (79.3%), followed by tactile (27.6%), taste (25.3%), visual (12.6%), and olfactory (5.7%) (Table 3).

Between the two groups, there were no significant differences in gender, receipt of public assistance, absence of a parent, or IQ (Table 4). The mean age of the HS group was significantly lower than that of the non-HS group. Regarding the CBCL T scores those of Total, Internalizing,

Withdrawn, and Anxious/Depressed of the HS group were significantly higher than those of the non-HS group.

Based on these results, we selected age, gender, PDD subtypes, and T scores of Withdrawn and Anxious/Depressed on the CBCL as independent variables for logistic regression analyses (Table 5). Our results showed that autistic disorder (OR=2.71, 95% CI=1.11-6.61, p=0.03) and a relatively elevated T score of Anxious/Depressed (OR=1.06, 95% CI=1.00-1.13, p=0.04) were significantly associated with sensory hypersensitivity.

Table 1. Characteristics of subjects

	Total subjects n=132
Age; years	10.7 (2.7)
Male /Female	101 (76.5%) /31 (23.5%)
IQ	96.0 (12.5)
Socioeconomic status	
Receipt of public assistance	5 (3.8%)
Absence of a parent	37 (28.0%)
Subtypes of PDD	
Autistic disorder	97 (73.5%)
Asperger' s disorder	2 (1.5%)
PDD-NOS	33 (25.0%)

Values are expressed as mean (SD) or n (%). IQ, Intelligence quotient; PDD, Pervasive developmental disorder; and PDD-NOS, Pervasive developmental disorder - not otherwise specified.

Table 2. Number of domains with sensory hypersensitivity of subjects

	n	%
none	45	34.1
only one area	55	41.7
two	21	15.9
three	10	7.6
four	1	0.8
five	0	0

The sum of n is equal to the total number of subjects (n=132). Values are n and %.

Table 3. Five domains of children with sensory hypersensitivity

	n	%
Auditory	69	79.3
Tactile	24	27.6
Olfactory	22	25.3
Visual	11	12.6
Gustatory	5	5.7

The sum of n is not equal to the total number of children with sensory hypersensitivity (n=87), because some children had sensory hypersensitivity in multiple domains.

Discussion

To our knowledge, this is the first study to report a correlation between the HFPDD subtype and sensory hypersensitivity. Sensory modulation symptoms including sensory hypersensitivity have been reported to associate with PDD from the time that autism has been defined as a diagnosis[34]. Although many studies have assessed the incidence of sensory hypersensitivity in children with PDD, there is no consensus as to whether hypersensitivity should be considered as core features of PDD.

In this study, after controlling for age, gender, IQ, socioeconomic status (absence of a parent; receipt of public assistance), and broadband psychopathology, children with autistic disorder that was

most severe subtype were more likely to exhibit sensory hypersensitivity than those with other PDD subtypes. These findings suggest that sensory hypersensitivity is associated with core features of PDD.

In the present study, 67% of the children with HFPDD displayed sensory hypersensitivity, in accordance with many other studies that have reported unusual sensory responses in the majority of children with PDD. Although some researchers have reported a prevalence rate of sensory hypersensitivity of 56%-70% in children with PDD[10,35], these rates were relatively higher than those

Table 4. Characteristics and CBCL scores between sensory hypersensitivity group and sensory non-hypersensitivity group

	Total	Subgroups		Analysis
	HFPDD (n=132)	HS group (n=87)	non-HS group (n=45)	p
Demographic variables				
Age; years	10.7 (9.1)	10.3 (2.7)	11.5 (2.5)	0.009 [a]*
Male/Female, n (%)	101(76.5)/31(23.5)	63(72.4) /24 (27.6)	38 (84.4) /7 (15.6)	0.122 [c]
Socioeconomic status				
Receipt of public assistance, n (%)	5 (3.8)	5 (5.7)	0 (0.0)	0.165 [c]
Absence of a parent, n (%)	37 (28.0)	25 (28.7)	12.0 (26.7)	0.802 [c]
IQ, mean (SD)	96.0 (12.5)	98.5 (12.2)	94.7 (12.6)	0.143 [a]
Autistic disorder, n (%)	97 (73.5)	71.0 (81.6)	26.0 (57.8)	0.003 [c]*
CBCL, mean (SD)				
Total	70 (8.8)	71.4 (9.1)	67.4 (7.6)	0.014 [a]*
Internalizing	67.8 (10.1)	69.8 (10.5)	63.7 (7.8)	0.000 [a]*
Externalizing	66.5 (10.8)	66.9 (10.5)	65.6 (11.5)	0.528 [a]
Withdrawn	67.5 (10.6)	69.3 (11.0)	64.0 (8.7)	0.011 [b]*
Somatic Complaints	59.6 (9.6)	60.4 (10.0)	58.1 (8.8)	0.245 [b]
Anxious/Depressed	66.6 (9.8)	68.6 (10.1)	62.4 (7.8)	0.000 [a]*
Social Problems	68.1 (9.7)	68.9 (9.8)	66.6 (9.4)	0.199 [a]
Thought Problems	64.8 (11.5)	66.1 (11.4)	62.2 (11.3)	0.087 [b]
Attention Problems	68.5 (8.1)	69.3 (8.4)	67.0 (7.2)	0.118 [a]
Delinquent Behaviour	63.2 (9.1)	63.2 (8.8)	63.0 (9.8)	0.964 [b]
Aggressive Behaviour	65.9 (10.3)	66.3 (10.1)	65.1 (10.8)	0.399 [b]

HFPDD, High-functioning pervasive developmental disorder; HS, Sensory hypersensitivity; IQ, Intelligence quotient; and CBCL, Child Behavior Checklist. Values are expressed as mean (SD) or n (%). [a] *t*-test. [b] Mann-Whitney *U*-test. [c] chi-square analysis. *$p<0.05$.

Table 5. Multiple logistic regression analysis for sensory hypersensitivity

Variable	Odds Ratio	95% CI	p
Age	0.88	0.75-1.03	0.11
Gender, Male	1.86	0.60-5.82	0.29
Autistic disorder	2.71	1.10-6.61	0.03*
T scores of CBCL			
Withdrawn	1.01	0.96-1.06	0.73
Anxious/Depressed	1.06	1.00-1.13	0.04*

Odds ratios >1 indicates higher likelihood of sensory hypersensitivity; values <1 indicate no likelihood of sensory hypersensitivity. CBCL, Child Behavior Checklist. *$p<0.05$.

shown in the general population (10%-17%)[20,35]. Importantly, these estimates did not exclude children with mental retardation. Nonetheless, despite the different cultural backgrounds and different intellectual levels, as with the present study, the aforementioned studies found a high prevalence of sensory hypersensitivity associated with PDD[10,35]. Overall, these findings suggest that sensory hypersensitivity is highly prevalent in PDD cases regardless of IQ.

In this study, sensory hypersensitivity was most commonly associated with auditory , which was consistent with the findings of previous studies[7,8]. Furthermore, Leekam et al[7] indicated that children with PDD had sensory abnormalities across multiple sensory domains, which was in accordance with our results that also showed that children with HFPDD tended to exhibit sensory hypersensitivity not only in one sensory domain but also in two or more.

Although age, gender, and IQ differences in sensory hypersensitivity of children with HFPDD have not been fully investigated, a recent study by Lai et al[36] reported that adult females with HFPDD frequently exhibited more lifelong sensory symptoms than males. In contrast to this study, we found no significant association between sensory hypersensitivity and gender, which may be explained by the fact that our subjects were children; therefore, further longitudinal studies are required. In the present study, age and IQ were not significantly associated with sensory hypersensitivity, as in accordance with the findings of Rogers et al[9], which showed that sensory hypersensitivity was unrelated to overall mental age or IQ in autistic children or those with developmental delays, in contrast to the authors' expectations.

In the present study, we found a significant correlation between sensory hypersensitivity and anxiety and/or depression. Some studies reported that sensory abnormalities were directly associated with anxiety and depression[6,17,26,27]. Tsuji et al[26] reported that children with HFPDD with sensory hypersensitivity exhibited more serious psychopathologies, particularly internalizing symptoms such as anxiety and depression. Although previous studies suggested that sensory hypersensitivity in children with PDD was associated with anxiety, they included children with mental retardation[6,27]. In the present study, after controlling for all other variables, including broadband psychopathology, we found significant associations to HFPDD, suggesting that sensory hypersensitivity in HFPDD was not associated with any psychopathology other than anxiety and depression. However, the causal relationship between anxiety and depression symptoms and the incidence of sensory hypersensitivity remains unclear.

The limitations of this study included the reliability and validity of the criteria for sensory hypersensitivity because they were not yet sufficiently established. In addition, we did not measure the severity of sensory hypersensitivity. Therefore, in future studies, operational criteria or a rating scale to measure the severity of sensory hypersensitivity should be established. Furthermore, the assessment of broadband psychopathology was based on parental reports. Although potential biases inherent in single-informant studies exist, parental reporting may provide a more valid indicator of functional abilities rather than self-reporting by children with HFPDD. Even among individuals with functional language skills, children and adults afflicted with HFPDD often have difficulty with emotional insight and expression[37]. Hence, the use of parental reporting for assessment of anxiety and other psychopathologies is recommended, although more precise results may be obtained by direct observation of the child.

In conclusion, our findings suggested that sensory hypersensitivity may be a core symptom of HFPDD and related to the incidence of anxiety and depression compared with other psychopathologies.

Although sensory hypersensitivity may significantly interfere with daily functioning, children with HFPDD may be not able to verbalize their sensory hypersensitivity symptoms because of impaired communication skills; thus, the number of children experiencing sensory hypersensitivity may be underestimated. Accordingly, it is important to assess sensory hypersensitivity at the time of diagnosis, and clinicians should minutely examine sensory hypersensitivity in HFPDD children with autistic disorder or with coexistent symptoms of anxiety and depression. Thus, the aggressive assessment of sensory hypersensitivity in children with PDD by clinicians can encourage support and ameliorate the quality of life.

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