

Sex differences in tactile defensiveness in children with ADHD and their siblings

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Tactile defensiveness (TD) is a disturbance in sensory processing and is observed in some children with attention-deficit-hyperactivity disorder (ADHD). TD has been examined in male children with ADHD and in children with ADHD without differentiating by sex. As males and females with ADHD may differ in the clinical expression of the disorder and associated deficits, the aim of this study was to examine sex differences in TD in males and females with ADHD. Non-affected siblings were also examined to investigate familiarity of TD. The Touch Inventory for Elementary-School-Aged Children was administered to 47 children with ADHD (35 males, 12 females; mean age 9y 8mo [SD 1y 11mo]), 36 non-affected siblings (21 males, 15 females; mean age 8y 10mo [SD 2y 4mo]), and 35 control children (16 males, 19 females; mean age 9y 5mo [SD 6mo]). Results indicated that females with ADHD displayed higher levels of TD than males with ADHD (who did not differ from control males). This suggests that TD is sex specific and may contribute to the identification of ADHD in females, thus improving diagnostic and therapeutic strength in this under-referred group. Non-affected siblings were unimpaired, regardless of sex, which suggests that TD is specific to the disorder and not part of a familial risk for ADHD.

Attention-deficit-hyperactivity disorder (ADHD)¹ is a common neuropsychiatric developmental disorder, which is estimated to affect between 3 and 11% of children and adolescents.² The core clinical symptoms of ADHD (inattention, hyperactivity, and impulsivity) are often accompanied by associated symptoms such as sleep problems, reduced motor coordination, and impaired balance.²

Sex possibly influences the clinical manifestation of ADHD and the extent to which associated symptoms of ADHD occur. Three studies found that females with ADHD showed relatively lower levels of hyperactivity and fewer diagnoses of conduct disorder and other comorbid disruptive behavior problems compared with males with ADHD.³⁻⁵ However, it was also found that females with ADHD have greater intellectual impairment,^{4,5} a higher prevalence of the predominantly inattentive subtype of ADHD,³ and are more at risk for substance-use disorders.³ One study found no sex difference with respect to impulsivity, academic performance, social functioning, and/or fine motor skills.⁴ Further study may help to determine whether differences exist between males and females with ADHD.

Relatively little is known about sex differences in disorders associated with ADHD, such as sensory processing disorders. Recent literature suggests that children with ADHD show a disturbance in sensory processing and integration of tactile stimuli.⁶⁻⁹ One area of dysfunction in sensory processing disorders is called tactile defensiveness (TD).^{10,11} TD is often observed in children with autism, learning disability,* fragile X syndrome, emotional disturbances, and specific learning difficulties.¹⁰⁻¹⁴ Individuals with TD show a tendency to 'react negatively and emotionally to certain touch situations'.¹⁴ They avoid touch and interpret non-noxious touch, such as light, brief touch to the arms, face, or legs, as threatening. They may try to avoid certain textures or types of clothing. Children with TD show an aversion to having their hair combed, nails cut by others, and avoid activities with a clear tactile component, such as finger painting.¹⁵

Four studies showed that children with ADHD exhibited higher levels of TD than control children.⁶⁻⁹ However, three of these studies did not differentiate between males and females in the data analyses⁷⁻⁹ and one study included only males.⁶ As sex may differentially affect the clinical manifestation of ADHD,^{3,4} and as a recent twin study showed that relatively more females than males have extreme TD scores, the main goal of the present study was to assess TD in males and females with ADHD to examine whether levels of TD are comparable in both sexes.

A second aim of this study was to assess TD in non-affected siblings of children with ADHD to examine whether TD is a familial characteristic of the disorder. The reason for including non-affected siblings alongside affected children is that this allows for investigation of a possible shared aetiology of TD and ADHD. That is, non-affected siblings share part of the (genetic and environmental) risk factors that have contributed to ADHD in their affected sibling. If TD arises partly from the same risk factors as ADHD, then one would expect to observe some TD in non-affected siblings, even in the absence of ADHD. If, however, ADHD and TD do not arise from the same risk factors but are merely associated with each other on a phenotypical level, then it is more likely that TD would be observed only in affected children, but not in familiarly at-risk siblings.

See end of paper for list of abbreviations.

*North American usage: mental retardation.

Method

PARTICIPANTS

Children with ADHD and their non-affected siblings participated at the Amsterdam site of the International Multicenter ADHD Genes (IMAGE) study.¹⁶ The IMAGE project is an international collaborative study which aims to identify genes that increase the risk of ADHD. Families with at least one child with ADHD and at least one additional sibling (regardless of possible ADHD status) were recruited from 12 specialist clinics in eight European countries. At the Amsterdam site, 190 families agreed to participate of which 178 families fulfilled all the criteria. Exclusion criteria were an IQ <70, diagnosis of autism, epilepsy, general learning difficulties (i.e. severe problems in multiple areas of academic learning), brain disorders, or known genetic disorders, such as Down syndrome or fragile X syndrome. All children were of European Caucasian descent.

This study was initiated as a pilot study on a subsample of the 178 families. Data collected during a 6-month period of recruitment of the families were available for 47 children (mean age 9y 8mo, SD 1y 11mo) with the combined subtype of ADHD (35 males: seven were not on medication, 25 took stimulants, one took non-stimulants, two took a combination of stimulants and non-stimulants; and 12 females: four were not on medication, six took stimulants, two took non-stimulants) and 36 non-affected siblings (21 males, 15 females; mean age 8y 10mo, SD 2y 4mo). An additional 35 control children (16 males, 19 females; mean age 9y 5mo, SD 6mo) were recruited from elementary schools in the same geographical region as the participating families of the children with ADHD. Only children aged between 6 and 12 years were included. All children were off medication for at least 48 hours (stimulants) or longer (non-stimulants) before testing. The study was approved by the local medical ethics committee and consent/assent forms were signed by parents and children of 12 years of age.

Within an affected family, the child clinically diagnosed with ADHD and the child's sibling were similarly screened using the standard procedures of the IMAGE project.^{16,17} To

identify children with ADHD symptoms, the Conners' ADHD Rating Scale (parent and teacher long versions),¹⁸ and the parent and teacher Strength and Difficulties Questionnaires (SDQ)¹⁹ were administered. Children with T-scores ≥ 63 on the Conners' ADHD Total scale (*Diagnostic and Statistical Manual of Mental Disorders*, 4th edn¹, Total symptom score) and scores >90th centile on the SDQ hyperactivity scale were considered as clinical. Siblings were regarded as non-affected if their scores were in the non-clinical range on both of the parent and teacher questionnaires (Conners' ADHD Total scale: T-score ≤ 62 , SDQ <90th centile). Subsequently, a semi-structured, standardized, investigator-based interview, the Parental Account of Children's Symptoms (PACS),²⁰ was administered for each child scoring clinically on any of the questionnaires completed by parents or teachers. No PACS interview was undertaken for non-affected siblings. The Conners' Scale (long version) for parents and teachers was completed for control children. Control children had to obtain non-clinical scores on both the parent and teacher versions (Conners' ADHD Total scale: T-score ≤ 62 ; see Table I).

TACTILE DEFENSIVENESS

The Touch Inventory for Elementary-School-Aged Children (TIE)¹⁵ was used to obtain a measure of TD. The TIE is a screening questionnaire for school-aged children aged between 6 and 12 years. It contains 26 items that involve common daily situations in which a child meets touch or stimulation. Children were presented with questions such as: 'Does it bother you to have your face washed?' and 'Does it bother you to go barefooted?' They were required to rate these situations on a 3-point scale in terms of the level of inconvenience: 'no', 'a little', 'a lot'. The TIE has a good internal reliability (a coefficient alpha of 0.79 and standardized alpha 0.79),¹⁵ test-retest reliability ($r=0.91$, $p<0.001$),²¹ and validity (discriminant analysis: $p=0.007$, 85% correct classification).¹⁵

The English TIE was translated into Dutch for the current study using double translation procedures (i.e. from English to Dutch and back to English again by separate individuals

Table I: Participants' attention-deficit-hyperactivity disorder screening scores by group

| | ADHD group | | Non-affected siblings | | Controls | | $F_{5,112}$ | Contrasts ($p<0.05$) |
|----------------------------------|-------------|-------------|-----------------------|-------------|------------|-------------|-------------|------------------------|
| | Males | Females | Males | Females | Males | Females | | |
| | (n=35) | (n=12) | (n=21) | (n=15) | (n=16) | (n=19) | | |
| Age, y:mo | 10:0 (1:7) | 10:0 (1:1) | 9:0 (2:2) | 8:11 (2:2) | 9:7 (0:6) | 9:4 (0:8) | 2.0 | ns |
| Conners' Scale – Parent | | | | | | | | |
| DSM-IV inattentive | 70.5 (7.8) | 78.8 (12.9) | 48.6 (9.7) | 48.2 (7.4) | 48.3 (8.4) | 44.9 (10.6) | 43.4 | 1=2 > 3=4=5=6 |
| DSM-IV hyperactive/ impulsive | 77.2 (8.8) | 78.2 (15.7) | 50.7 (9.5) | 49.7 (7.6) | 50.1 (9.7) | 48.2 (4.5) | 50.6 | 1=2 > 3=4=5=6 |
| DSM-IV Total | 75.5 (7.3) | 80.6 (12.6) | 49.3 (9.5) | 48.9 (7.1) | 49.1 (8.5) | 47.6 (3.6) | 68.2 | 1=2 > 3=4=5=6 |
| Anxious-shy | 59.6 (13.3) | 63.6 (16.4) | 50.3 (10.7) | 50.1 (10.2) | 48.9 (8.3) | 50.7 (10.5) | 4.7 | 1=2, 1>5, 2>3=4=5=6 |
| Conners' Scale – Teacher | | | | | | | | |
| DSM-IV inattentive | 63.4 (5.7) | 72.4 (11.3) | 47.0 (5.3) | 51.0 (5.6) | 46.9 (5.9) | 45.7 (2.0) | 56.7 | 2>1 > 3=4=5=6 |
| DSM-IV hyperactive/ impulsive | 68.1 (9.2) | 75.4 (9.7) | 49.9 (5.9) | 49.8 (6.0) | 45.0 (2.7) | 45.3 (1.7) | 64.5 | 2>1 > 3=4=5=6 |
| DSM-IV Total | 67.1 (6.7) | 75.8 (10.1) | 48.3 (5.6) | 50.5 (4.3) | 45.7 (4.3) | 44.7 (2.2) | 84.9 | 2>1 > 3=4=5=6 |
| Anxious-shy | 61.6 (11.0) | 60.7 (13.4) | 56.1 (8.6) | 53.5 (11.0) | 53.2 (9.1) | 57.9 (13.4) | 2.1 | ns |

All values are mean (SD) unless otherwise indicated. ADHD, attention-deficit-hyperactivity disorder. DSM-IV, *Diagnostic and Statistical Manual of Mental Disorders*, 4th edn.¹ Contrasts: 1, males with ADHD; 2, females with ADHD; 3, non-affected male siblings; 4, non-affected female siblings; 5, control males; 6, control females; ns, not significant.

who were fluent in English and Dutch). Any discrepancies were resolved after mutual agreement. A previous study has shown that mean test scores are similar in North American children (English version of the TIE) and European children (non-English version of the TIE translated into the native language of the children),²² suggesting that careful translation of the TIE does not affect results. Scores could hypothetically range between 26 and 78, but the typical range is between 26 and 60 based on a sample of 415 children.²² The same study showed that the mean score of the TIE was 41 with an SD of 7.8. Based on these data, a score of 57 and above ($\geq 2SDs$) was regarded as a clinical score for TD. Scores of the TIE are referred to as TD scores.

DATA ANALYSIS

Analyses were performed using SPSS (version 14.1). A mixed model was used with sex (two groups: males and females) and group (three groups: ADHD, non-affected siblings, and controls) as fixed factors. Family was used as random effect to account for within-family correlation, as more than one child per family participated resulting in dependency of observations. The 'sex by group' interaction was implemented in the model to investigate whether sex differences on the TIE were similar between groups. Using 2SDs as a cut-off (raw score ≥ 57), the percentage of children with extreme scores on the TIE was calculated for each group.

Results

A mixed model with sex and group as fixed factors, the 'sex by group' interaction, and family as a random effect, showed no main effect of sex ($F[1, 117.8]=0.97, p=0.33$), a significant main effect of group ($F[2, 107.6]=3.51, p=0.03$), and a significant 'sex by group' interaction ($F[2, 117.3]=4.14, p=0.02$). The latter finding implied that both main effects (sex and group) could not be interpreted separately and analyses should be separated by group. When analyses were rerun, females had significantly higher TD scores than males with ADHD ($F[1, 47.0]=10.27, p=0.002$) and control females ($F[1, 31.0]=13.06, p=0.001$). Males with ADHD did not differ from control males ($F[1, 48.0]=0.01, p=0.91$). No sex difference was found when non-affected siblings were analyzed ($F[1, 32.1]=0.16, p=0.70$) or when controls were analyzed ($F[1, 35.0]=0.59, p=0.45$), and non-affected siblings did not differ from controls ($F[1, 49.8]=0.29, p=0.59$; see Fig. 1).

Using a cut-off score of 2SD extreme TD scores were found in: one male with ADHD, two females with ADHD, one non-affected male sibling, one non-affected female sibling, one control male, but no control female.

Discussion

The aim of the present study was to examine whether TD is comparable in males and females with ADHD, and whether TD is also present in non-affected siblings of children with ADHD.

Overall, children with ADHD had higher TD scores than control children, which is concordant with previous studies.⁶⁻⁹ These findings highlight the presence of deficits associated with ADHD outside the spectrum of the core symptoms of inattention, hyperactivity, and impulsivity, and may suggest underlying abnormalities in the processing of somatosensory stimuli. Optimally-functioning sensory processing enables an adequate range of performance and adaptation to the daily environment.⁸ In young children, poor sensory processing

may cause difficulties in their social, cognitive, and sensory-motor development.⁹ Moreover, problems in sensory processing may lead to difficulties in originating adequate responses in school, home, and community settings.⁷ Examining sensory processing in children with ADHD might prove valuable for intervention purposes.

The findings indicate that levels of TD were different according to sex: females with ADHD portrayed more TD than males with ADHD. Moreover, the group of males with ADHD did not differ from the group of control males, whereas the group of females with ADHD differed from the group of control females. When the percentage of extreme-scoring cases was calculated, about 17% of females with ADHD had extreme scores (possibly indicating TD), whereas only 3% of males with ADHD had extreme scores. In contrast to a previous study of males with ADHD⁶, the current study did not find elevated levels of TD in the group of males with ADHD. An explanation for this might be that the present study and the study of Parush et al.⁶ differed in method of TD measurement. Parush et al. used the Touch Inventory for Preschoolers which is based on teacher reporting, whereas this study used the TIE based on self-report. As three of four previous studies on TD in ADHD did not take the effect of sex into account,⁷⁻⁹ the current finding of sex differences for TD in ADHD cannot be compared with those previous studies.

There might be several explanations for the difference in TD between males and females with ADHD. Perhaps the most obvious explanation would be that females in general have higher levels of TD than males, so that the sex differences found in the current study are not unique to ADHD. Some evidence for this was found in a recent twin study¹⁰ in which proportionally more females than males had extreme TD scores. However, in the same twin study it was reported that there was no difference between groups of males and females for mean TD scores. This is concordant with the current finding of an

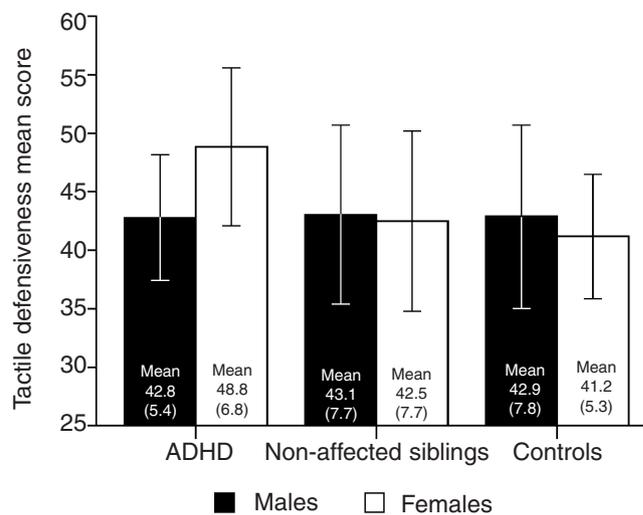


Figure 1: Mean (SD) scores of Tactile defensiveness (TD) in children with attention-deficit-hyperactivity disorder (ADHD), their non-affected siblings, and controls for males and females separately. Compared with other groups, females with ADHD showed higher levels of TD.

insignificant main effect of sex on TD scores. Thus, it appears that the sex differences for TD found in this study are specific to children with ADHD and are not related to general sex differences for TD.

A second explanation for the elevated TD in females with ADHD compared with males with ADHD might be found in the relationship between TD, anxiety, and inattention. The inattentive subtype of ADHD is relatively more frequently diagnosed in females than in males³ and the inattentive subtype is more strongly associated with internalizing problems, such as anxiety, than the combined ADHD subtype.⁵ Recently, it was found that TD correlated with a fearful temperament and anxiety.¹⁰ Perhaps TD is an 'anxious' response to certain tactile situations, expressed in emotional responses to, or avoidance of, non-noxious touch and stimuli that are interpreted as threatening.^{14,15} To examine whether inattention, anxiety, and TD were interrelated in the current sample, correlations between these measures were calculated, and all were significant.^a Furthermore, females with ADHD differed more clearly from controls in their increased levels of anxiety (reported by parents) and inattention (reported by teachers) than males with ADHD. These findings support the explanation that a triad of symptoms (TD, anxiety, and inattention) co-occurs and that these symptoms appear more severe in females than in males with ADHD.

A third explanation for the increased levels of TD in females compared with males with ADHD might be specifically related to the sample of females with ADHD. It is known that fewer females with ADHD are diagnosed and referred for treatment than males with ADHD,³ which may suggest that the females included in this study had a more severe form of ADHD than the males. This might translate into more severe levels of TD in females than in males. The current sample of females had higher scores on both the inattentive and hyperactivity/impulsivity scales (rated by teachers) than the sample of males with ADHD.

Furthermore, TD and anxiety did not appear to be uniquely associated with symptoms of inattention (see second explanation), but were also associated with symptoms of hyperactivity/impulsivity;^b this suggests that the elevated TD in females compared with males with ADHD might be related to a more severe form of general pathology in the females of this sample. This is concordant with previous findings of correlations between severity of psychopathology and TD.⁸ However, the explanation of a generally more severe pathology in females compared with males with ADHD in the current sample is contradicted by the equal raw scores of inattention and hyperactivity/impulsivity between both ADHD groups, suggesting comparable severity of ADHD symptoms in absolute terms between both ADHD groups. Therefore, future research should seek to determine whether increased levels of TD in females compared with males with ADHD relate to higher levels of inattention and/or higher levels of overall psychopathology in females compared with males.

This appears to be the first study reporting on TD in non-affected siblings of children with ADHD to examine the

familiarity of TD. The group of non-affected siblings (males and females) had normal TD scores, which may suggest that TD found in ADHD is associated with the disorder at a phenotypical level but does not (wholly) relate to the same familial risk factors associated with ADHD.¹⁷ However, it may also indicate certain causal factors not shared by affected and non-affected siblings. The finding that non-affected siblings had normal TD scores may be important for future research into the causal pathways leading up to ADHD, and to determine why certain children develop ADHD while other (at-risk) children do not.

There are some limitations to interpretation of the current findings. The overall sample size, and particularly the size of the group of females with ADHD, was relatively small. Future research using larger groups would help to clarify whether the current results can be replicated. Furthermore, only one (screening) measure of TD was used, and that measure was based on self-report. It is recommended that future studies apply a larger variety of TD measures, preferably also more experimental measures of TD to examine altered processing and/or appraisal of tactile stimuli.

Another limitation that should be noted is that even though the English version of the TIE was translated very carefully into the Dutch language, the Dutch translation of the TIE has not been investigated for validity and reliability, which should be carried out in future studies using this Dutch translation. However, this is unlikely to have influenced the results in the current study, as previous research has shown that mean test scores were similar in North American children (English version of the TIE) and European children (non-English version of the TIE translated into native language of the children).²² These studies suggest that careful translation of the TIE does not affect results.

Care should be taken when interpreting results based on group means. Although, overall, males with ADHD had TD scores comparable to those of control children, some extreme cases were present (just as in the non-affected sibling groups and control groups), suggesting that TD might be present in some males with ADHD, their non-affected siblings, and even in typically developing control children. These cases should not be overlooked.

Conclusion

The current findings indicate that more females than males with ADHD suffer from increased levels of TD. Assessment of TD may contribute to an increased identification of ADHD in females who tend to be under-diagnosed,³ enabling improved intervention. As non-affected siblings, both males and females, did not display TD, TD does not appear to be part of a familial risk for ADHD.

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^aAll correlations between TD, anxiety, and inattention were positive and ranged between 0.19 and 0.44 (all *p* values <0.05).

^bAll correlations between hyperactivity/impulsivity and TD and anxiety were positive and significant, ranging from 0.19 to 0.36 (all *p* values <0.05).

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List of abbreviations

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| IMAGE | International Multicenter ADHD Genes study |
| PACS | Parental Account of Children’s Symptoms |
| SDQ | Strengths and Difficulties Questionnaire |
| TD | Tactile defensiveness |
| TIE | Touch Inventory for Elementary-School-Aged Children |

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