

ADHD Symptoms and School Impairment History in Parents of ADHD Children are a Fundamental Diagnostic and Therapeutic Clue.

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Research

Keywords: Attention Deficit Hyperactivity Disorder, Familiarity, Wender Utah Rating Scale-25, School impairment, Emotional lability

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TITLE PAGE

Title: ADHD symptoms and school impairment history in parents of ADHD children are a fundamental diagnostic and therapeutic clue.

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ABSTRACT

Background: Attention Deficit and Hyperactivity Disorder (ADHD) is a multi-factorial condition, with inheritance playing a major role. Recognizing parents' ADHD represents a clue not only for an earlier diagnosis of the disease in their children, but also to optimize psycho-educational therapy outcome, by addressing the impairment of parenting related to untreated ADHD. This study aimed to assess the frequency of features suggestive of ADHD during childhood among parents of affected children, and the presence of school and emotional impairment.

Methods: We administered the Wender Utah Rating Scale-25, a self-assessment tool for the retrospective identification of symptoms consistent with ADHD during childhood, to a cohort of 120 parents of 60 children diagnosed with ADHD, and to a consistent number of "controls".

Results: The WURS-25 proved positive in 49.1% of fathers and 30.0% of mothers of ADHD patients, compared to 1.7% of fathers and 1.7% of mothers of non-ADHD patients ($p < 0.0001$).

The questions addressing learning and emotional impairment provided significantly higher scores in parents with an overall positive test compared to those with negative test ($p < 0.0001$).

Conclusions: This study demonstrates a remarkably high rate of symptoms consistent with ADHD during childhood in parents of affected children. Physicians should be aware that this is a relevant anamnestic clue and, given the relevance of parents' role in the management of children with ADHD, an important issue in order to improve patients' treatment.

Keywords: Attention Deficit Hyperactivity Disorder; Familiarity; Wender Utah Rating Scale-25; School impairment; Emotional lability

TEXT

Background

Attention Deficit and Hyperactivity Disorder (ADHD) is a condition characterized by marked, persistent, maladaptive levels of inattention, impulsiveness, and hyperactivity, which has a negative impact on social, educational, and professional performances. Its estimated prevalence worldwide is 5% in children and 2.5% in adults¹, with studies showing that in half the cases, the disorder persists during adulthood^{2,3}. According to a survey⁴, 40% of children diagnosed with ADHD undergo remission during adulthood, in 40% of cases symptoms persist in an attenuated form with related emotional deregulation, social and professional difficulties, while 20% continue to show features consistent with the typical form of the disease.

Although not yet fully understood, ADHD aetiology is probably multi-factorial. Several factors have been demonstrated to be involved, including low birth weight, smoking, drug and alcohol exposure during pregnancy, among biological factors⁵ and maternal psychiatric disorders, family dysfunction and lower socioeconomic status among environmental modifiers⁶. The identification of specific causative genes is still hampered by the significant phenotypic heterogeneity of the disorder⁷. Nevertheless, the role of inheritance has been well clarified⁸: twin concordance amounts to 70-76%^{9,10,11} and the presence of an affected first-degree relative has been demonstrated to give four times higher risk, to any child, to develop the disorder¹².

This study aimed to assess the frequency of features suggestive of ADHD during childhood among parents of affected children, and the presence of school difficulties and emotional lability.

Methods

A prospective case-control study was performed at the Child Neurology and Psychiatry Unit of the University teaching, tertiary children's hospital, Institute for Maternal and Child Health "Burlo Garofolo", in Trieste, Italy, from April 2019 to October 2019. The study was approved by the Institutional Review Board and all participants gave their written consent to take part in it.

We considered as “cases” the parents of children, who had received a diagnosis of ADHD (of any of the following types: impulsive/hyperactive, inattentive and distractible, combined) between January 2005 and June 2019. The control group consisted of parents of children with various neuropsychiatric conditions, summarized in *Figure 1*, and did not include those of patients who presented other conditions leading to attention or behavioural deficits, potentially acting as confounding factors. Furthermore, since all ADHD patients had an intellectual quotient (IQ) over 70, we selected comparable controls, thus excluding also parents of patients with $IQ \leq 70$.

We initially identified 202 patients; 142 were not included due to the presence of exclusion criteria or to the impossibility to contact their families. Sixty ADHD children (56 males) were therefore finally considered, along with 120 parents (60 mothers and 60 fathers): the latter were considered as “cases” and were matched to 60 mothers and 60 fathers of non-ADHD children as a control group.

Regarding the disease subtypes, 2/60 (3.3 %) children had predominant hyperactivity, 14/60 (23.3%) predominant inattention, and 44/60 (74.3%) a combined disorder. The 44/60 (73%) of children had at least one co-morbidity: 35/60 (58%) of patients displayed Oppositional Defiant Disorder, 11/60 (23%) Specific Learning Disorder, 10/60 (17%) Mood or Anxiety Disorder, and 7/60 (12%) other disorders, such as mixed learning disorder, language disorder, autism, epilepsy or obsessive-compulsive disorder, and 17/60 (28%) of enrolled patients had more than one co-morbidity. Finally, 46/60 (77%) of children with ADHD were on ongoing drug therapy with methylphenidate, in 15/60 (25%) in association with an antipsychotic drug, with one patient taking risperidone as a single drug; 13/60 (22%) were not receiving any drug therapy.

The Wender Utah Rating Scale-25 (WURS-25)^{13,14}, in the Italian language (*Figure 2*), was administered online to all participating parents. The WURS-25 is a self-assessment tool for the retrospective identification of the presence and severity of symptoms consistent with ADHD during childhood (age 6–10 years). It consists of 25 items, of which 21 address ADHD (inattention, hyperactivity, impulsivity, affective, emotional, and functional dysfunction), and four serve as control items. This tool has shown good psychometric properties and satisfactory internal and temporal reliability, and it is considered a screening tool for the retrospective assessment of ADHD in childhood. It has been demonstrated that ADHD symptoms undergo remission through adulthood in a relevant percentage of the cases. Therefore, when

assessing the presence of symptoms suggestive of ADHD in the adult population, there is a substantial risk of underestimating the real prevalence of the disorder in childhood, making WURS a more suitable tool for this study, providing an accurate picture of the period in which the disorder may have been more expressed.

We instructed the subjects to recall their behaviour and mood during primary school (age 6 to 10), rating every item from 0 to 4 (not at all or very slightly (score=0), mildly (score=1), moderately (score=2), quite a bit (score=3), or very much (score=4)). The total score range from 0 to 100: we considered 46 as the cut-off score suggestive for previous ADHD.

Four questions (number 22-25) aimed to evaluate scholastic impairment, while seven questions (number 2-3, 9, 11-13, 17) concerned the presence of emotional lability.

We used the Chi-Square Test for the dichotomous variables and the Fisher exact test in case of frequencies below 5. For continuous variables, we used the Wilcoxon-Mann Whitney test (for all distributions, the Kolmogorov-Smirnov test for normality had a p-value < 0.05).

Results with p-value ≤ 0.05 were considered statistically significant.

Results

Among parents of ADHD children, 60/60 mothers (100%) and 57/60 fathers (95%) answered the questionnaire, versus 60/60 (100%) and 60/60 (100%) mothers and fathers of non-ADHD children, respectively.

The test proved positive (score ≥ 46) in 46/117 parents of children with ADHD (39.3%) and 2/120 (1.7%) parents of non-ADHD children ($\chi^2 = 51.99$, $p < 0.0001$) (Figure 3).

Among parents of ADHD children, we detected a higher rate of positivity in fathers (28/57, 49.1%) compared to mothers (18/60, 30.0%) ($\chi^2 = 4.48$, $p = 0.034$), while this difference was not observed among controls (1/60, 1.7% fathers and 1/60, 1.7% mothers; $\chi^2 = 0.000$, $p = 1.000$).

As for mothers, 94.7% of the positive tests belonged to "the case group", which meant that they had a child with ADHD ($\chi^2 = 17.01$, $p < 0.0001$). Similar results were observed in fathers, with a slightly higher correlation (96.6% vs. 3.4%, $\chi^2 = 29.43$, $p < 0.0001$).

In parents tested positive, a concomitant compromise of the school and emotional functioning was sought. The questions, aimed at investigating school impairment, provide a score ranging from 0 to 16 points. The median score was 8 (IQR 7-10) among the 48 parents with overall WURS positive test, versus 3 (IQR 1-4) among the 189 parents testing negative (*Table 1, Figure 3*). Of interest, 28.3% of parents with overall WURS positive test, repeated at least one class. Through the Wilcoxon-Mann-Whitney test, we compared the scores regarding school impairment of parents with an overall positive and negative WURS test, finding significantly higher scores among parents with a positive test ($p < 0.0001$). The median score was 9 (IQR 7-12) among the 19 mothers who tested positive, against 3 (IQR 1-4) among the 101, who were negative. Statistically a significant difference was found between the two groups ($p < 0.0001$). Besides, 26.3% of mothers with positive tests reported having repeated at least one class during the school period. The median score of fathers was 8 (IQR 7-9) among the 29 who tested positive, versus 3 (IQR 2-5) among the 88 who tested negative. Even in this case, there was a statistically significant difference between the two groups ($p < 0.0001$), and again, the 27.6% of fathers with positive tests reported having repeated at least one class during the school's years.

The questions related to the presence of emotional lability, provide a total score between 0 and 28 points. The median score was 14 (IQR 11.5-15) among the 48 parents with positive tests, versus 5 (IQR 3-7) among the 189, who were negative. The difference was statistically significant ($p < 0.0001$) (*Table 2, Figure 3*). The median score of mothers was 14 (IQR 14-15) among the 19 who were positive, versus 5 (IQR 3-7) among the 101, who tested negative ($p < 0.0001$). For fathers, the median score was 13 (IQR 10-15) among the 29 who were positive, versus 5 (IQR 3-7) among the 88, who proved to be negative ($p < 0.0001$).

Discussion

This study shows a high rate of symptoms consistent with ADHD during childhood in parents of children affected by this disease.

Several aspects are involved in the multi-factorial pathogenesis of ADHD, including genetic, neurobiological and environmental elements. Inheritance probably plays a fundamental role in pathogenesis: the assessment of its weight would allow not only a better understanding of the disease but also an early diagnosis in the children of affected parents, along with timely treatment and prevention of detrimental consequences. Remarkably, a positive family history may be a relevant diagnostic clue, which physicians should specifically address. On the other hand, parent training is a cornerstone of treatment for children with mild-to-moderate ADHD¹⁵. Since parental ADHD may particularly impair parenting and family functioning, hindering the ability to deal with affected children, identifying affected parents could be a clue to optimize patients' outcomes^{16,17}.

In this study we found a significant correlation between kinship and parents' WURS test positivity: in particular, 49.1% of fathers and 30.0% of mothers of ADHD patients, compared to 1.7% of fathers and 1.7% of mothers of non-ADHD patients, had a positive test, presenting features consistent with ADHD during their childhood. Remarkably none of the analyzed parents had ever been diagnosed with ADHD, underlying a lack of standardized diagnostic criteria to detect this condition in the past. However, the WURS test does not allow the retrospective diagnosis of ADHD: indeed, its functionality is to highlight the presence of emotional and behavioural traits consistent with the disease. These results are in line with the previous literature. The role of genetic factors has been assessed over time⁸, and it has been confirmed by the high concordance between twins^{3,9,11}. Bidwell demonstrated a four-time higher risk of developing ADHD in children with affected parents or first-degree relatives, compared to the general population¹². Starck and colleagues found a WURS test positivity in 49.1% of fathers and 27.3% of mothers of ADHD patients¹⁸. Compared to the survey mentioned above, our study was powered by the analysis of data from a control group and by the comparable number of enrolled fathers and mothers, which allowed us to stratify the

weight of familiarity by the sex of affected parents. While previous studies relied on the WURS test to assess symptoms suggestive of ADHD during childhood in parents of affected children, to our knowledge, this is the first study that separately examines the school and emotional impairment of parents during their childhood, through a specific sub-analysis of the scores obtained in the specific test questions, addressing these issues.

Regarding school performances, we found a correlation between reported parents' school difficulties and WURS test positivity, confirming the pivotal role of undiagnosed and untreated ADHD in learning problems. The latter, as underlined by Marzocchi and colleagues⁷, can be explained in the light of a vicious circle between the deficit of self-regulation cognitive processes and the inability to adopt effective organizational and executive strategies appropriate to the task. This profile negatively affects the performance in the comprehension of written texts, studying, and resolution of arithmetic problems^{19,20}. In this study, we found that mothers with positive tests showed higher scores in the questions investigating school impairment, compared to fathers, whose tests were positive. This issue could further demonstrate that in females with ADHD, inattention is usually preponderant compared to hyperactivity, negatively affecting academic performances. Similar results were found regarding emotional deregulation. Parents with an overall positive WURS test demonstrated higher scores in the questions related to emotional lability, compared to parents with negative tests.

Tabassam and Grainger widely described the emotional dysfunction of ADHD children, characterized by sudden emotional changes, dysphoria, irritability, low tolerance to frustration, emotional hyper-reactivity, and emotions-recognition deficits²¹. The concurrent effect of the critical judgment of families, teachers, and peers, easily led to low self-esteem, sense of social rejection and loneliness, which in turn can promote the development of further psychopathologies, such as mood disorders. By stratifying data by gender, we found that the mothers tested positive presented a median score higher compared to fathers tested positive, underlying the central role of emotional lability within the disorder in females. In a longitudinal study involving 140 children diagnosed with ADHD, Hinshaw found that female subjects had more severe anxiety, depressive symptoms, and more significant difficulties in daily functioning than males²². At ten

years of follow-up, girls with ADHD were more likely to exhibit self-injuring behaviors and suicide attempts than boys.

This study, not only confirms these data but also highlights a high occurrence of school and emotional impairment in parents of affected children during their childhood, strengthening the suitability of the WURS test for the retrospective assessment of symptoms suggestive of ADHD, in light of the crucial role of these two aspects in the diagnosis of the disorder.

The main limits of this study consist of the relatively small sample size and the lack of correlation analysis between patients and parents, due to the choice to collect and investigate the results in a completely anonymous way. For the same reason, we assessed every parent independently; therefore, it was not possible to compare the correlation with an affected child of a single positive parent and both parents' positivity. On the other hand, the guarantee of anonymity provided a parental participation rate of almost 100%, which in turn, represents the strength of this study. A further point of strength was the case-control design, which allowed additional comparative results.

Conclusions

This study demonstrated a high rate of symptoms consistent with ADHD during childhood in parents of children affected by this pathology. Physicians should actively investigate parents' history when evaluating children with a suspected ADHD syndrome. A proper assessment of the parents of these patients would also be crucial to optimize psycho-educational results.

List of abbreviations: ADHD, Attention Deficit and Hyperactivity Disorder; IQ, Intellective Quotient; WURS-25, Wender Utah Rating Scale-25; IQR, Interquartile Range

DECLARATIONS

Ethics approval and consent to participate: The study was approved by the Institutional Review Board of the Institute for Maternal and Child Health – IRRCCS “Burlo Garofolo”, where it was conducted, and all participants gave their written consent to take part in it.

Consent for publication: Patients and/or their caregivers gave written consent for data publication.

Availability of data and materials: All data generated or analysed during this study are included in this published article.

Competing interests: The authors declare that they have no competing interests.

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Authors' contributions: AS designed the study, GP collected the data; VR performed the statistical analysis; LCW and FR drafted the work, and EB edited the manuscript; all the authors approved the final version of the manuscript and take full responsibility for its contents.

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TABLES

Table 1. Parents stratified by study group and WURS-25 test result (positive/negative).

	Parents of ADHD children			Parents of non-ADHD children			p-value
	N.	Negative test	Positive test	N.	Negative test	Positive test	
Total	117	71 (60.7%)	46 (39.3%)	120	118 (98.3%)	2 (1.7%)	<0.0001
Mothers	60	42 (70%)	18 (30%)	60	59 (98.3%)	1 (1.7%)	<0.0001
Fathers	57	29 (50.9%)	28 (49.1%)	60	59 (98.3%)	1 (1.7%)	<0.0001

Table 2. Median and interquartile range (in parenthesis) score of questions assessing school difficulties and emotional functioning stratified by sex with Wilcoxon-Mann-Whitney test p-values.

		Positive		Negative		p-value
		N.	Median	N.	Median	
School difficulties	Mothers	19	9.0 (9)	101	3.0 (8)	< 0.0001
	Fathers	29	8.00 (9)	88	3.00 (9)	< 0.0001
	Total	48	8.0 (9)	189	3.0 (9)	< 0.0001
Emotional deregulation	Mothers	19	14.0 (5)	101	5.0 (14)	< 0.0001
	Fathers	29	13.0 (8)	88	5.0 (15)	< 0.0001
	Total	48	14.0 (8)	189	5.0 (15)	< 0.0001

FIGURE LEGEND

Figure 1. Co-morbidities in non-ADHD patients

Figure 2. The Wender Utah Rating Scale (WURS) 25

Figure 3. Scores distribution of the WURS-25 test comparing cases and controls, and on questions investigating school impairment and emotional deregulation comparing subjects with an overall positive or negative test.

Figures

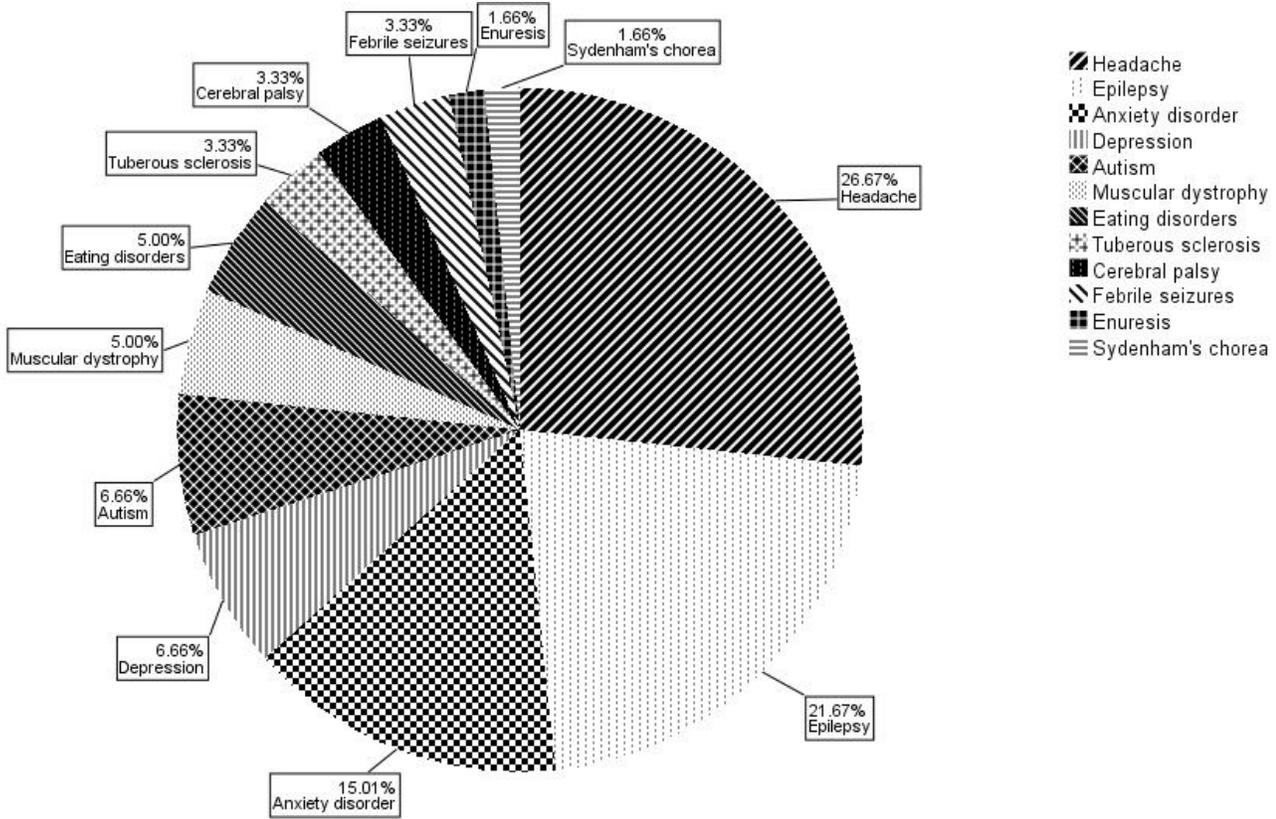


Figure 1

Co-morbidities in non-ADHD patients

- As a child I was (or had):

		0	1	2	3	4
1	Easily distracted					
2	Anxious worrying					
3	Nervous fidgety					
4	Inattentive daydreaming					
5	Hot- or short-tempered					
6	Temper outbursts tantrums					
7	Trouble not finishing things					
8	Stubborn strong-willed					
9	Sad or blue depressed					
10	Disobedient with parents					
11	Low opinion of myself					
12	Irritable					
13	Moody ups and downs					
14	Angry					
15	Impulsive, acting without thinking					
16	Tendency to be immature					
17	Guilty feelings regretful					
18	Losing control of myself					
19	Tendency to be or act irrationally					
20	Unpopular, not very friendly					
21	Trouble seeing someone else's view					
22	Trouble with authorities					
23	Overall a poor student					
24	Troubles with math or numbers					
25	Not achieving up to potential					

Where: 0 = Not at all or slightly; 1 = Mildly; 2 = Moderately; 3 = Quite a bit; 4 = Very much

- Has you repeated classes? (indicate which)

Figure 2

The Wender Utah Rating Scale (WURS) 25

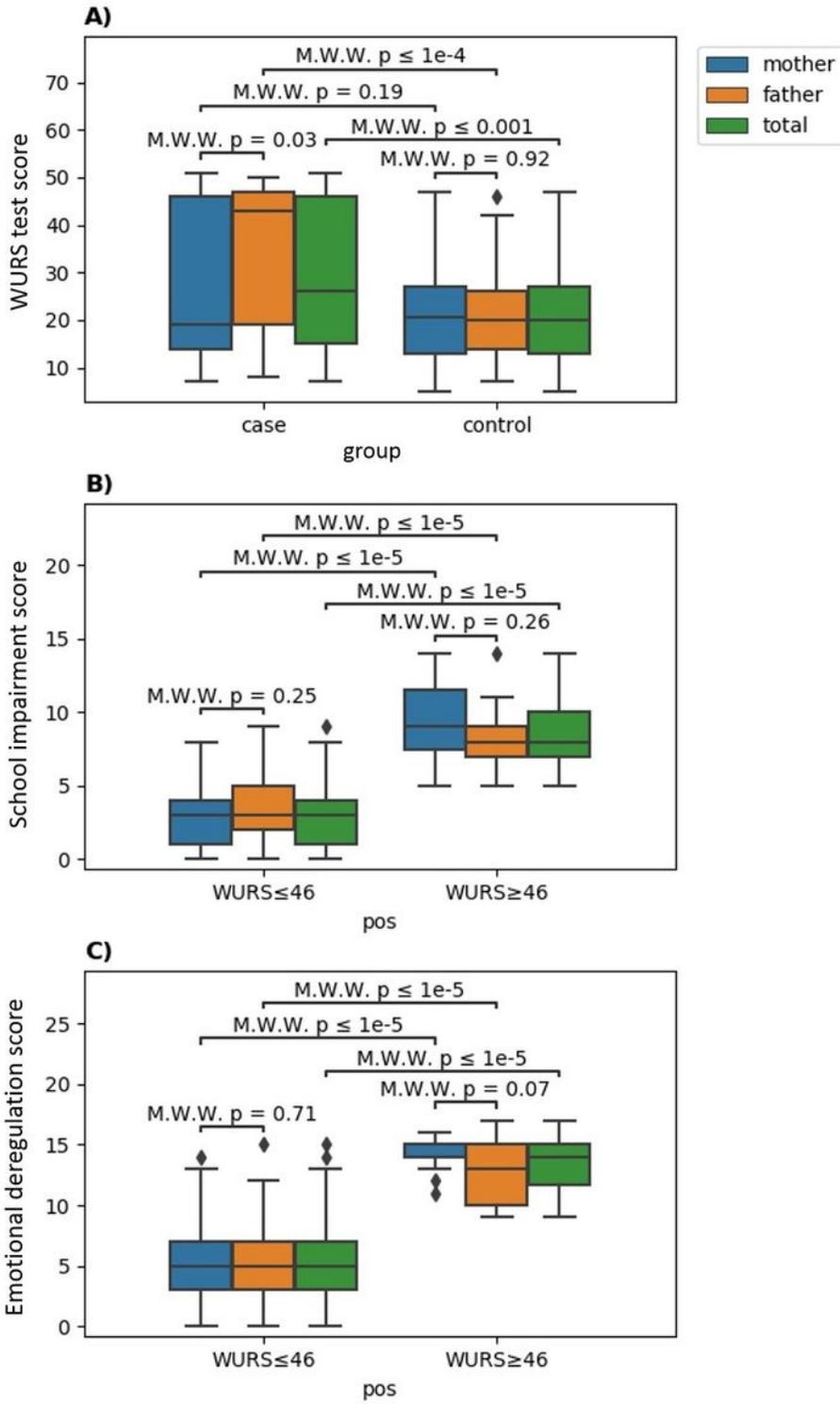


Figure 3

Scores distribution of the WURS-25 test comparing cases and controls, and on questions investigating school impairment and emotional deregulation comparing subjects with an overall positive or negative test.