

The behavioural profile of children with attention-deficit/hyperactivity disorder and of their siblings

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Abstract The behavioural profiles in $N = 69$ index children with attention-deficit/hyperactivity disorder (ADHD), $N = 32$ siblings with ADHD, $N = 35$ siblings without ADHD, and $N = 36$ normal controls were compared by the use of standardized parent and teacher rating scales. The four groups were matched by age and IQ. The behavioural profiles of the two ADHD groups were very similar not only in the behavioural domains of ADHD, but also in scales measuring emotional and conduct problems. Siblings without ADHD shared more similarities with normal controls except for more emotional problems. These general trends were stronger in the parent compared to the teacher ratings. These findings indicate that not only ADHD-related but also other behaviours show a strong family aggregation. The informant

differences may reflect context dependent differences in child behaviour and contrast effects particularly in parental ratings.

Keywords Attention-deficit/hyperactivity disorder · Behaviour · Siblings

Introduction

High concordance of attention-deficit/hyperactivity disorder (ADHD) diagnoses between child and parents were found both in early and more recent familial aggregation studies [1–6]. The risk for ADHD is two- to eightfold in parents, and also elevated in siblings of children with ADHD [7]. In addition, a higher rate of antisocial and affective disorders has also been shown in the relatives of ADHD children [8–12]. The association with antisocial disorders in relatives is particularly strong for children with combined ADHD and conduct disorder [9, 12].

Findings in siblings of children with ADHD show an intermediate level of affection between affected probands and unaffected controls with respect to ADHD symptoms [7] and comorbid conditions [13]. The heightened psychopathological risk for siblings of children with ADHD has also been shown in a longitudinal study. At a 4-year follow-up, significant elevations of behavioural, mood, and anxiety disorders were found among the siblings of ADHD children [14].

In contrast to these family aggregation studies based on structured interviews resulting in clinical diagnoses, there are only a few studies dealing with the behaviour of siblings of ADHD children. Faraone et al. [14] used the Child Behavior Checklist [15] and found evidence of psychosocial dysfunction that aggregated among the siblings with

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ADHD. However, the study by Faraone et al. [14] did not include index children (patients) with ADHD but, rather, concentrated on siblings of children with ADHD and controls only both at baseline and at a 4-year follow-up.

In a more recent study by Listug-Lunde et al. [16], only parent-report measures indicated that siblings of children with ADHD had higher levels of internalizing and ADHD-related behaviours than control siblings, whereas child-report measures did not show any significant group differences. Similar to the study by Faraone et al. [14], also the study by Listug-Lunde et al. [16] did not include ADHD patients themselves and reported only a few aggregated scores of questionnaires but no detailed behavioural profiles. Both studies did not contain teacher information.

The large International Multi-centre ADHD Genetics (IMAGE) project has collected behavioural data based on the Conners' Questionnaires [17] and the Strength and Difficulties Questionnaire [18] from 1,068 probands with ADHD and 1,446 unselected siblings. This study found larger differences between probands and siblings at home than at school in most of the behavioural scores reflecting contrast effects in the parents [19, 20]. In another study from this project, the clinical correlates and familial prevalence of emotional lability in children and adolescents with ADHD have been assessed [21].

Within the Multi-level Family Assessment of ADHD project (MFAA) including behavioural, neuropsychological, neurophysiological, and genetic assessments in ADHD children, one of their siblings, and their parents, we have been following the dimensional approach of behavioural assessment in the various family members, along with quantitative neuropsychological and neurophysiological assessments [22, 23]. The present contribution had the following aims: (1) to test the hypothesis that the similarity of ADHD Index children and their siblings will largely be due to shared ADHD, (2) to test the hypothesis that siblings without ADHD will not be completely free from any behavioural abnormalities but more similar to control children, and (3) to test the impact of parent and teacher questionnaires based on the hypothesis that observed behavioural differences between groups will also depend on the informant.

Methods

Samples

The recruitment included both referred and non-referred participants. The referred children came from a public child and adolescent psychiatric service and from local paediatricians and child and adolescent psychiatrists in private practice. Furthermore, participants came from a

large national ADHD self-help group or responded to various campaigns in the media providing information on the project. Inclusion criteria of the clinical group were: (a) at least one child with ADHD combined type, (b) participation of two children and both parents in the study, (c) children living with both parents, (d) children aged 5–17 years, (e) European origin, and (f) sufficient knowledge of German in both children and parents. Furthermore, autism in the index child served as an exclusion criterion. Control families had to fulfil the criteria (b)–(f). No attempt was made to draw a representative sample in terms of the distribution of socioeconomic status.

A total of 172 children participated in the study and their parents gave informed consent. The participating ADHD families consisted of both biological parents and of two children (except two families with a single index child only) aged 8–16 years, with at least one index child meeting criteria for DSM-IV combined type and no selective criteria for the siblings. Based on specific ADHD assessment procedures, there were 69 ADHD Index children (mean age = 11.4, SD = 2.0 years; m:f ratio 3:1; mean IQ = 115.9, SD = 16.5), 32 siblings with ADHD (mean age 11.3, SD = 2.7 years; m:f ratio 0.7:1; mean IQ = 117.2, SD = 18.3), and 35 siblings without ADHD (mean age 11.7, SD = 2.5 years; m:f ratio 0.52:1; mean IQ = 115.9, SD = 15.1). Whereas fulfilment of criteria for ADHD combined type was mandatory for the index children, ADHD subtypes varied in the affected siblings. The latter comprised 9 combined, 19 inattentive, and 4 hyperactive-impulsive subtypes. The 36 control children (mean age 11.1, SD 2.1 years; m:f ratio 1.25:1; mean IQ = 119.5, SD = 16.5) were recruited from regional elementary school, friends, or local sports club. There were no significant age differences ($F = 0.58$, $df = 3$, NS) and no significant IQ differences ($F = 0.43$, $df = 3$, NS) between the four samples.

Assessments

Rating scales used to quantify ADHD symptoms included the German versions of the Conners' Parent Rating Scale [17], the Conners' Teacher Rating Scale [17], the Strengths and Difficulties Questionnaire, parent and teacher version [18], and the Child Behavior Checklist [15]. The various primary and secondary scales of these instruments may be seen from the tables in the "Results" section. Parents and teachers were asked to rate the behaviour of the child when the child had been off medication. *T*-scale scores based on the US-American standardization samples were used for the CPRS and the CTRS. The SDQ analyses are based on raw scores because there is no standardization and the CBCL data are based on the local Zurich standardization [24]. All scales show sufficient reliability coefficients in the various standardization samples.

The Parental Account of Children's Symptoms (PACS) [25], a semi-structured, standardized, investigator-based interview was used for the diagnostic assessment of ADHD. Children with their families were included if at least one child met criteria of the DSM-IV combined type, as resulting from both the PACS and items that scored 2 and 3 from the teacher-rated Conners' ADHD Index of the CTRS. The detailed diagnostic procedure and algorithm (HYPESHEME) has been described in an early paper coming from the IMAGE project [26]. The present study was in part modelled after the IMAGE project [19, 20]. DSM-IV subtypes of siblings were defined according to the HYPESHEME algorithm, with the exception of three siblings. In these cases, no PACS interview was conducted and siblings were classified according to the concurrent CPRS and CTRS *L*- and/or *M*-scores ($T > 60$).

For control children, CTRS, CPRS, SDQ, and CBCL were completed, and non-clinical scores were required for inclusion. To control for intelligence, the following subtests of the Wechsler Intelligence test for children (WISC-III; Wechsler) [27] were assessed: vocabulary, similarities, block design, and picture completion. The intelligence quotient (IQ) was prorated from these subtests using an algorithm developed by Schallberger [28].

Statistical analyses

Two parallel statistical procedures were performed when comparing the findings in the four samples. To control for unequal sample sizes, normal distribution of variables (analyzed with the Kolmogorov–Smirnov-test), homogeneity of variances (Levene test), and homogeneity of variance–covariance matrices (Box's *M* test) were checked first. With a few exceptions, in most of the variables there was a violation of these prerequisites of the analysis of variance model. As a consequence, the non-parametric Kruskal–Wallis test was performed as a first strategy of data analysis.

In a second approach, group comparisons were performed by the use of multivariate analyses of covariance controlling for age and sex (MANCOVA). If these analyses did not explain more variance than multivariate analyses of variance without these co-variables (MANOVA), the latter were followed by Tamhane's *T*₂ post hoc tests for unequal variances. Finally, if there were no differences in the level of significance between the non-parametric and the parametric approach, the MANOVA model was preferred because of better control of chance findings.

After controlling for the potential impact of the co-variables and after comparing both statistical approaches, the final data to be reported here are based only on MANOVA and MANCOVA models. All analyses were performed with the help of the Statistical Program for Social Sciences (SPSS, version 16.0).

Results

In the following, findings from group comparisons of index children with ADHD, their siblings with ADHD, siblings without ADHD, and normal controls will be described for the various questionnaires used in this study. Table 1 presents findings based on the CPRS. Because the three DSM-IV scales of the CPRS were used for diagnosis, they were not included in the analyses. As one can see, both multivariate comparisons of the seven primary scales (upper part of the table) and the three secondary scales (lower part of the table), and the univariate comparison of the total score revealed highly significant differences across the four samples. The post hoc tests indicate that with the exception of hyperactivity, there were no differences between index children with ADHD and their siblings with ADHD on any scale. Siblings without ADHD were similar to control children in the domains of oppositional behaviour, cognitive problems/inattention, hyperactivity, social problems, Conners' ADHD Index, and Conners' Global Index (CGI): Impulsive. However, siblings without ADHD scored significantly higher than normal controls on the scales measuring anxious/shy behaviour, perfectionism, CGI: Emotional Lability, and on the total score. With the exception of perfectionism and social problems, index children and their siblings with ADHD scored higher than siblings without ADHD and controls.

The parallel findings based on the SDQ parent version are presented in Table 2. Again, the MANOVA comparing the subscale scores was highly significant as was the ANOVA of the total scores. With the exception of the two scales measuring hyperactivity and the total score, there were no significant differences between index children and their siblings with ADHD. These two groups scored significantly higher than siblings without ADHD and controls on all scales except on prosocial behaviour. The latter two groups did not differ significantly on any scale of the SDQ parent version.

Comparisons of the four groups based on the CBCL are presented in Table 3. Both MANOVA and ANOVA findings indicate that there were significant group differences on all levels of primary and secondary scales and the total score. On most of the primary and secondary scales, i.e. social withdrawal, somatic complaints, anxious/depressed, social problems, thought problems, delinquent behaviour, internalizing, and total scores, index children and siblings with ADHD scored equally high, whereas index children scored even higher than siblings with ADHD on inattention, aggressive behaviour, and externalizing problems. These two groups of children with ADHD scored higher than the two other groups without ADHD on various scales, namely, on inattention, delinquent behaviour, aggressive behaviour, externalizing, and total problems.

Table 1 Comparisons of the Conners' Parent Rating Scale (CPRS) findings in four groups of children

	Index children with ADHD (A)		Siblings with ADHD (B)		Siblings without ADHD (C)		Control children (D)		<i>F</i>	<i>P</i> (<i>df</i> = 3)	Post hoc comparisons
	Mean	SD	Mean	SD	Mean	SD	Mean	SD			
Oppositional behaviour	71.8	10.8	65.9	11.8	56.1	11.0	50.5	6.3	41.14	<0.001	A = B > C = D
Cognitive problems/inattention	71.8	70.3	71.1	12.3	51.6	9.6	48.9	6.6	65.56	<0.001	A = B > C = D
Hyperactivity	78.5	9.7	66.7	13.1	51.8	9.8	47.4	4.9	106.27	<0.001	A > B > C = D
Anxious/shy	59.7	12.5	62.2	15.2	52.6	10.8	46.6	6.8	13.95	<0.001	A = B > C > D
Perfectionism	61.6	10.2	60.5	11.1	54.8	11.0	48.4	6.1	16.27	<0.001	A = B > D; A > C; C > D
Social problems	67.5	14.9	62.9	14.2	54.9	11.2	50.1	7.4	17.54	<0.001	A = B > D; A > C
Psychosomatic	62.3	14.1	64.8	16.4	51.3	8.6	49.3	9.3	14.37	<0.001	A = B > C = D
Conners' ADHD Index	73.5	9.6	71.1	12.1	49.8	9.8	46.9	5.3	93.85	<0.001	A = B > C = D
CGI: Impulsivity	75.7	9.3	69.8	12.8	51.4	9.6	46.8	4.6	101.87	<0.001	A = B > C = D
CGI: Emotional Lability	66.9	12.4	62.8	13.0	52.1	10.4	46.7	5.7	32.78	<0.001	A = B > C > D
Conners' Global Index: Total	75.1	9.8	69.0	12.9	51.8	10.2	46.5	4.6	89.17	<0.001	A = B > C > D

Primary scales: Wilks' $\lambda = 0.252$, $F = 13.67$, $df = 21$; 466; $P < 0.001$

Secondary scales: Wilks' $\lambda = 0.321$, $F = 26.69$, $df = 9$; 404; $P < 0.001$

Total score: $F = 89.17$, $df = 3$; 168; $P < 0.001$ (ANOVA)

CGI Conners' Global Index

Table 2 Comparisons of parent-rated Strengths and Difficulties Questionnaire (SDQ) findings in four groups of children

	Index children with ADHD (A)		Siblings with ADHD (B)		Siblings without ADHD (C)		Control children (D)		<i>F</i>	<i>P</i> (<i>df</i> = 3)	Post hoc comparisons
	M	SD	M	SD	M	SD	M	SD			
Emotional problems	4.0	2.5	3.7	2.7	1.7	2.1	1.1	1.5	17.60	<0.001	A = B > C = D
Conduct problems	4.4	2.1	3.3	2.0	1.8	1.9	3.0	2.3	106.65	<0.001	A = B > C = D
Hyperactivity	7.9	1.8	5.8	2.6	2.1	2.2	1.7	1.4	30.62	<0.001	A > B > C = D
Peer problems	3.6	2.7	2.9	2.6	1.4	1.6	1.3	1.6	11.36	<0.001	A = B > C = D
Prosocial behaviour	6.7	2.4	7.0	2.3	7.8	1.9	8.4	1.4	5.83	0.001	A = B < D
Total score	19.9	5.9	15.7	6.8	7.0	5.6	5.1	3.5	73.03	<0.001	A > B > C = D

Primary scales: Wilks' $\lambda = 0.320$, $F = 15.32$, $df = 15$; 450; $P < 0.001$

Total score: $F = 73.03$, $df = 3$; 167; $P < 0.001$ (ANOVA)

On the other scales measuring somatic complaints, anxious/depressed, social problems, thought problems, and internalizing the differentiation between subjects with or without ADHD, respectively, was less precise with the exception that index children always scored higher than controls.

The comparisons of the four groups based on teacher ratings are presented in the next two tables. Findings on the CTRS are collected in Table 4 and are based on MANCOVA, because the inclusion of the co-variables age and sex was characterized by a markedly higher proportion of explained variance than the MANOVA model. The multivariate analyses indicate strong effects of group, sex, and

age on all levels of the questionnaires. The post hoc group comparisons show that the two groups with ADHD scored higher than the other two groups without ADHD on the scales measuring cognitive problems/inattention, Conners' ADHD Index, and CGI: Impulsive. It should be noted that the scores of the four groups on the CTRS ADHD Index are in accordance with the diagnostic algorithm using items with scores 2 and 3 as part of the classification. Because the DSM-IV-based items of ADHD of the CTRS were both represented in the diagnostic algorithm and in the Conners' ADHD Index as a dependent variable, the differentiation by this variable simply reflects the diagnostic definition of the groups. In addition, index children scored higher than

Table 3 Comparisons of parent-rated Child Behavior Checklist (CBCL) findings in four groups of children

	Index children with ADHD (A)		Siblings with ADHD (B)		Siblings without ADHD (C)		Control children (D)		<i>F</i>	<i>P</i> (<i>df</i> = 3)	Post hoc comparisons
	M	SD	M	SD	M	SD	M	SD			
Social withdrawal	58.4	8.4	59.4	10.1	56.4	7.5	52.4	5.0	5.36	0.002	A = B, > D, C = D
Somatic complaints	58.3	7.3	57.5	7.9	55.6	7.1	53.6	6.6	3.53	0.016	A = B=C, A > D
Anxious/depressed	60.1	8.5	58.9	9.9	55.2	6.3	51.9	4.6	9.52	<0.001	A = B, A > C, A > D, B > D
Social problems	62.5	7.8	60.7	8.1	56.6	7.9	53.5	4.9	13.10	<0.001	A = B, A > C, A > D, B > D
Thought problems	56.4	8.8	54.6	7.8	52.9	6.0	51.5	4.3	3.84	0.011	A = B = C; A > D
Inattention	67.6	7.0	62.9	7.5	54.4	7.6	51.6	3.6	55.00	<0.001	A > B > C = D
Delinquent behaviour	61.6	8.6	59.6	7.6	53.2	4.8	52.0	3.3	20.42	<0.001	A = B > C = D
Aggressive behaviour	67.2	10.1	60.7	8.6	54.1	6.3	52.0	3.7	34.68	<0.001	A > B > C = D
Internalizing	59.0	8.6	57.2	12.1	52.8	10.4	45.4	9.8	15.01	<0.001	A = B > D, A > C
Externalizing	66.3	10.0	59.5	11.1	50.5	9.5	46.2	8.5	39.28	<0.001	A > B > C = D
Total score	65.6	8.7	61.2	11.0	52.1	10.9	44.6	9.8	39.23	<0.001	A = B > C > D

Primary scales: Wilks' $\lambda = 0.408$, $F = 6.93$, $df = 24$; 458; $P < 0.001$
 Secondary scales: Wilks' $\lambda = 0.558$, $F = 18.50$, $df = 6$; 328; $P < 0.001$
 Total score: $F = 39.23$, $df = 3$; 165; $P < 0.001$ (ANOVA)

Table 4 Comparisons of Conners' Teacher Rating Scale (CTRS) findings in four groups of children

	Index children with ADHD (A)		Siblings with ADHD (B)		Siblings without ADHD (C)		Control children (D)		<i>F</i>	<i>P</i> (<i>df</i> = 3)	Post hoc comparisons
	M	SD	M	SD	M	SD	M	SD			
Oppositional behaviour	63.1	12.1	53.2	10.2	58.4	11.6	52.3	11.5	6.26	<0.001	A = B = C = D
Cognitive problems/ inattention	60.3	9.6	60.4	11.0	52.9	10.0	49.8	6.9	14.01	<0.001	A = B > C = D
Hyperactivity	67.5	11.4	61.1	13.6	51.5	7.0	50.6	7.8	19.16	<0.001	A > B > C = D
Anxious/shy	62.2	10.3	60.1	7.9	58.2	10.9	55.6	8.8	2.91	0.015	A = B = C, A > D
Perfectionism	59.1	8.6	57.3	10.2	53.4	8.5	54.1	8.9	3.77	0.003	A = B; A > C, A > D
Social problems	57.7	11.7	53.5	8.7	51.2	9.3	50.0	6.7	3.61	0.004	A = B; A > C; A > D
Conners' ADHD Index	69.0	9.8	63.2	11.2	53.0	9.4	49.0	6.6	34.17	<0.001	A = B > C = D
CGI: Impulsivity	68.8	10.1	62.9	12.1	53.5	9.7	49.4	8.6	25.38	<0.001	A = B > C = D
CGI: Emotional Lability	64.0	13.3	56.7	10.2	53.7	11.6	54.1	12.7	5.74	<0.001	A > B = C = D
Conners' Global Index: Total	68.9	10.5	61.9	11.7	54.1	10.1	50.9	9.1	20.20	<0.001	A > B > C = D

Primary scales: Wilks' $\lambda = 0.568$, $F = 5.54$, $df = 18$; 450; $P < 0.001$ (group); Wilks' $\lambda = 0.568$, $F = 5.54$, $df = 18$; 450; $P < 0.001$ (sex); Wilks' $\lambda = 0.568$, $F = 5.54$, $df = 18$; 450; $P < 0.001$ (age)
 Secondary scales: Wilks' $\lambda = 0.502$, $F = 14.37$, $df = 9$; 394; $P < 0.001$ (group); Wilks' $\lambda = 0.568$, $F = 5.54$, $df = 18$; 450; $P < 0.001$ (sex); Wilks' $\lambda = 0.568$, $F = 5.54$, $df = 18$; 450; $P < 0.001$ (age)
 Total score: $F = 20.20$, $df = 5$; 164; $P < 0.001$ (ANOVA: corrected model)

their siblings with ADHD and the latter scored higher than the other two groups without ADHD on the scales measuring hyperactivity and CGI: Total. Furthermore, index children with ADHD scored higher than all the other three groups on the CGI: Emotional Lability scale with the scores of the latter being not significantly different. There

were no significant differences among the four groups on the scales measuring oppositional behaviour, and the differentiation of the four groups was less precise on the scales measuring anxious/shy behaviour, perfectionism, and social problems. On these scales, at least, index children scored significantly higher than controls.

Table 5 Comparisons of teacher-rated Strengths and Difficulties Questionnaire (SDQ) findings in four groups of children

	Index children with ADHD (A)		Siblings with ADHD (B)		Siblings without ADHD (C)		Control children (D)		<i>F</i>	<i>P</i> (<i>df</i> = 3)	Post hoc comparisons
	M	SD	M	SD	M	SD	M	SD			
Emotional problems	2.8	2.1	2.2	2.1	1.5	1.6	1.6	1.9	4.53	0.004	A = B, A > C, A > D
Conduct problems	2.9	2.1	1.9	1.7	1.1	1.8	0.9	1.4	12.61	<0.001	A = B, A > C, A > D
Hyperactivity	6.8	2.5	4.9	2.3	2.4	1.8	2.1	1.8	48.40	<0.001	A > B > C = D
Peer problems	2.7	2.4	1.5	1.7	1.5	1.7	1.6	1.8	4.58	0.004	A > B = C = D
Prosocial behaviour	5.5	2.4	6.9	2.2	6.7	2.6	7.1	2.2	5.21	0.002	A < B = C = D; A < D
Total score	15.2	5.1	10.5	5.6	6.4	4.6	6.7	6.3	38.60	<0.001	A > B > C = D

Primary scales: Wilks' $\lambda = 0.485$, $F = 8.88$, $df = 15$; 445; $P < 0.001$

Total score: $F = 38.60$, $df = 3$; 165; $P < 0.001$ (ANOVA)

The final group comparisons are based on the SDQ teacher version and are shown in Table 5. Again, the multivariate and univariate analyses show that there were significant group differences. Post hoc comparisons indicate that index children showed higher scores than both siblings without ADHD and controls on all scales except reverse findings for prosocial behaviour. Index children had even higher scores than their siblings with ADHD on scales measuring hyperactivity, problems with peers, and on the total score than their siblings with ADHD. The latter scored higher on hyperactivity and total score than siblings without ADHD and controls.

Discussion

In the present study, behavioural features of ADHD children, their siblings, and unaffected controls were compared. Behavioural features of ADHD and other domains as well were compared by the use of both parent and teacher questionnaires. The major finding of the present study is the expected similarity in parental behavioural ratings between index children with ADHD and their siblings, who also qualified for a diagnosis of ADHD. However, our findings indicate that this similarity is not restricted to behavioural features, which are specific to ADHD but also relates to other behavioural domains like emotional and conduct problems as shown by the findings based on various scales of the CPRS, the SDQ, and the CBCL.

There were only a few scales with higher scores for the index children than their siblings with ADHD, e.g. the hyperactivity scales in both the CPRS and the SDQ parent version and the inattention and the aggressive scales of the CBCL, indicating that these differences might have contributed to the parental decision of defining the index child for the study. Thus, in general, our first hypothesis of a strong similarity of index children and siblings with ADHD across a wide array of behavioural dimensions and parental

questionnaires was clearly supported. Furthermore, the at-risk status of siblings with ADHD for meaningful clinical impairment that was found by Faraone et al. [14] was also strongly supported by the present study.

There was also a trend for siblings without ADHD to show a behavioural pattern that was much closer to the behaviour of controls rather than the behaviour of their sibling with ADHD. However, a few exceptions are noticeable. Siblings without ADHD scored significantly higher than normal controls on scales measuring in particular emotional problems and the total score of the CPRS, and on the total score of the CBCL. Thus, our second hypothesis was also clearly supported by showing that in terms of the behaviour profile, siblings without ADHD, in general, are more similar to controls than to their siblings with ADHD, but may show also some indication of behavioural abnormalities. Their emotional abnormalities may result from increased family stress with a partial neglect of their needs by the parents, who direct most of their parental engagement on the more difficult child with ADHD. Similar observations have been made in the families of chronically ill and handicapped children [29]. However, given the high familial aggregation of affective disorders in children with ADHD [10], genetic segregation of emotional problems may also play a role. However, a recent study based on a larger sample did not find that emotional lability and ADHD co-segregate in families [21]. A definite decision on the contribution of shared family environment versus genetic transmission lies beyond this study which was not designed for such a differentiation.

In an additional set of analyses, we studied teacher ratings of the behaviour of the four groups. Both the companion instruments of the Conners' questionnaires and the SDQ were employed. The Teacher Rating Form (TRF; Achenbach) [15] as the companion instrument to the CBCL was simply not employed because we had been afraid of a decline in the motivation of the teachers to collaborate with the study due to an overload of questions and some repetition of questionnaire items. The findings based on the

CTRS indicate that, in general, the differentiation of the two groups with ADHD from the other two groups without ADHD worked better on the various ADHD subscales of the questionnaire, whereas the differentiation between the groups in all other behavioural domains was less pronounced. On the much shorter SDQ, teachers also rated much less behavioural differences than parents between the four groups. In particular, the differentiation of the two sibling groups became less evident. Thus, also our third hypothesis on the impact of the informant was also clearly supported. Informant differences have been also detected in the data-set of the IMAGE project [19, 20]. They were particularly pronounced in probands, but almost absent in siblings, leading to significant rater effects. However, these informant effects were present only for scales representing externalizing behaviour.

In general, informant differences may be interpreted in various ways. First, they may reflect real differences in the behaviour in various settings. Thus, observable behaviour like ADHD may show some variations at school and at home. However, it should be pervasively present because otherwise the diagnosis would not have been correct. The clear differentiation of the four groups on almost all ADHD-related scales by the teacher does indicate that the diagnostic process was correct and that the informant is of minor importance in this respect. The less clear differentiation in other behavioural domains and, most particularly, on scales measuring emotional problems may be explained by a lack of opportunity to observe these behaviours at school. In comparison to teachers, parents have much better and more frequent opportunities to observe also emotional problems in their children, so that there was a much better differentiation of the groups regarding behavioural domains other than ADHD by the parents in the present study. These findings differ from those by Müller et al. [19, 20], who found smaller informant effects in emotional problems.

However, besides variations in behaviour depending on the context also rater effects have to be taken into consideration. Parents may be particularly prone to a halo effect with overemphasizing also other behavioural abnormalities next to ADHD symptoms in their problematic children with the result of a more homogenous behaviour profile in these children and deemphasizing behavioural abnormalities in their children without ADHD. These contrast effects may be particularly strong if the parents themselves show some ADHD features. This issue will be studied in further analyses based on the data from our family project.

In the present sample, girls were clearly overrepresented among the siblings and most of them were diagnosed ADHD-inattentive subtype. In part, the elevated female rate among the affected siblings may be due to study selection criteria

including hyperactive symptoms which more often occur in boys. Therefore, in a family with both a boy and a girl affected by ADHD, it is more likely for the boy to be selected as an index child. Similarly, if only one child is affected, it is most probably the boy. Not surprisingly, in the present sample, girls were also overrepresented among the non-affected siblings. Although the occurrence of emotional problems does not depend on gender, it needs to be emphasized that the non-affected sibling is more likely to be a girl, whose symptoms of emotional distress are at risk to be overlooked.

In conclusion, the findings of the present study should alert clinicians to not only concentrate on the referred child but also on the siblings, to provide both with professional assessment and treatment, and to keep in mind the sizeable proportion of non-referred female siblings. Even in the absence of ADHD there may be other behavioural problems in the children that may need some intervention to also reduce the impact of potential stress in a family with one or more members with ADHD. Limitations of the present study include the mixed recruitment with both referred and non-referred participants and the lack of any self-report based assessments of the participating children. Furthermore, the varying sex ratios in the various samples in combination with the limited sample sizes prevented more detailed analyses of sex differences.

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Conflict of interest The authors have no conflicts of interest to declare.

References

- Morrison J, Stewart M (1971) A family study of the hyperactive child syndrome. *Biol Psychiatry* 3:189–195
- Cantwell D (1972) Psychiatric illness in families of hyperactive children. *Arch Gen Psychiatry* 27:414–423
- Biederman J, Faraone SV, Keenan K, Benjamin J, Krifcher B, Moore C, Sprich-Buckminster S, Ulaglia K, Jellinek MS, Steinhard R, Spencer T, Norman D, Loldny R, Kraus I, Perrin J, Keller MB, Tsuang MT (1992) Further evidence for family-genetic risk factors in attention deficit hyperactivity disorder. Patterns of comorbidity in probands and relatives psychiatrically and pediatrically referred samples. *Arch Gen Psychiatry* 49:728–738
- Biederman J, Faraone SV, Keenan K, Knee D, Tsuang MT (1990) Family-genetic and psychosocial risk factors in DSM-III attention deficit disorder. *J Am Acad Child Adolesc Psychiatry* 29: 526–533
- Faraone SV, Biederman J, Milberger S (1994) An exploratory study of ADHD among second degree relatives of ADHD children. *Biol Psychiatry* 35:398–402

6. Smalley SL, McGough JJ, Del'Homme M, NewDelman J, Gordon E, Kim T, Liu A, McCracken JT (2000) Familial clustering of symptoms and disruptive behaviours in multiplex families with ADHD. *J Am Acad Child Adolesc Psychiatry* 39:1135–1143
7. Faraone SV (2004) Genetics of adult attention-deficit/hyperactivity disorder. *Psychiatr Clin North Am* 27:303–321
8. Biederman J, Faraone SV, Keenan K, Tsuang MT (1991) Evidence of familial association between attention deficit disorder and major affective disorders. *Arch Gen Psychiatry* 48:633–642
9. Biederman J, Munik K, Knee D (1987) Conduct and oppositional disorders in clinically referred children with attention deficit disorder: a controlled family study. *J Am Acad Child Adolesc Psychiatry* 26:724–727
10. Faraone SV, Biederman J (1997) Do attention deficit hyperactivity disorder and major depression share familial risk factors? *J Nerv Ment Dis* 185:533–541
11. Frick PJ, Lahey BB, Christ MAG, Loeber R, Green S (1991) History of childhood behavior in biological relatives of boys with attention-deficit hyperactivity disorder and conduct disorder. *J Clin Child Psychol* 20:445–451
12. Lahey B, Piacentini J, McBurnett M, Stone P, Hartdegen SE, Hynd G (1988) Psychopathology in the parents of children with conduct disorder and hyperactivity. *J Am Acad Child Adolesc Psychiatry* 27:163–170
13. Faraone SV, Biederman J, Keenan K, Tsuang MT (1991) Separation of DSM-III attention deficit disorder and conduct disorder: evidence from a family-genetic study of American child psychiatric patients. *Psychol Med* 21:109–121
14. Faraone SV, Biederman J, Mennin D, Gershon J, Tsuang M (1996) A prospective four-year follow-up study of children at risk for ADHD: psychiatric, neuropsychological, and psychosocial outcome. *J Am Acad Child Adolesc Psychiatry* 35:1449–1459
15. Achenbach TM (1991) Manual for the child behavior checklist/4–18 and 1991 profile/manual for the teacher rating form and 1991 profile. University of Vermont, Department of Psychiatry, Burlington
16. Listug-Lunde L, Zevenbergen AA, Petros TV (2008) Psychological symptomatology in siblings of children with ADHD. *J Atten Disord* 12:239–247
17. Conners CK (1997) Conners' Parent Rating Scale-Revised (L)/Conners' Teacher Rating Scale-Revised (L). Multi-Health Systems Inc., New York
18. Goodman R (1997) The Strengths and Difficulties Questionnaire: a research note. *J Child Psychol Psychiatry* 38:581–586
19. Müller UC, Asherson P, Banaschewski T, Buitelaar J, Ebstein R, Eisenberg J, Gill M, Manor I, Miranda A, Oades RD, Roeyers H, Rothenberger A, Sergeant J, Sonuga-Barke E, Thompson M, Faraone SV, Steinhausen HC (2011) The impact of study design and diagnostic approach in a large multi-centre ADHD study. Part 1: ADHD symptom patterns. *BMC Psychiatry* 11:54
20. Müller UC, Asherson P, Banaschewski T, Buitelaar J, Ebstein R, Eisenberg J, Gill M, Manor I, Miranda A, Oades RD, Roeyers H, Rothenberger A, Sergeant J, Sonuga-Barke E, Thompson M, Faraone SV, Steinhausen HC (2011) The impact of study design and diagnostic approach in a large multi-centre ADHD study. Part 2: dimensional measures of psychopathology and intelligence. *BMC Psychiatry* 11:55
21. Sobanski E, Banaschewski T, Asherson P, Buitelaar J, Chen W, Franke B, Holtmann M, Krumm B, Sergeant J, Sonuga-Barke E, Stringaris A, Taylor E, Anney R, Ebstein R, Gill M, Miranda A, Mulas F, Oades RD, Roeyers H, Rothenberger A, Steinhausen H-C, Faraone SV (2010) Emotional lability in children and adolescents with attention-deficit/hyperactivity disorder (ADHD): clinical correlates and family prevalence. *J Child Psychol Psychiatry* 51:915–923
22. Valko L, Doehner M, Müller UC, Schneider G, Albrecht B, Drechsler R, Maechler M, Steinhausen H-C, Brandeis D (2009) Differences in neurophysiological markers of inhibitory and temporal processing deficits in children and adults with ADHD. *J Psychophysiol* 23:235–246
23. Valko L, Schneider G, Doehner M, Müller UC, Brandeis D, Steinhausen H-C, Drechsler R (2010) Time processing in children and adults with ADHD. *J Neural Transm* 117:1213–1228
24. Steinhausen HC, Winkler Metzke C, Maier M, Kannenberg R (1997) Behavioral and emotional problems reported by parents for ages 6 to 17 in a Swiss epidemiological study. *Eur Child Adolesc Psychiatry* 6:136–141
25. Taylor E, Schachar R, Thorley G, Wieselberg M (1986) Conduct disorder and hyperactivity: I. Separation of hyperactivity and antisocial conduct in British child psychiatric patients. *Br J Psychiatry* 149:760–767
26. Brookes K, Xu X, Chen W, Zhou K, Neale B, Lowe N, Anney R, Franke B, Gill M, Ebstein R, Buitelaar J, Sham P, Campbell D, Knight J, Andreou J, Altink M, Arnold R, Boer F, Buschgens C, Butler L, Christiansen H, Feldman L, Fleischman K, Fliers E, Howe-Forbes R, Goldfarb A, Heise A, Gabriëls I, Korn-Lubetzki I, Marco R, Medad S, Minderaa R, Mulas F, Müller U, Mulligan A, Rabin K, Rommelse N, Sethna V, Sorohan V, Uebel H, Psychogiou L, Weeks A, Barrett R, Craig I, Banaschewski T, Sonuga-Barke E, Eisenberg J, Kuntsi J, Manor I, McGuffin P, Miranda A, Oades RD, Plomin R, Roeyers H, Rothenberger A, Sergeant J, Steinhausen H-C, Taylor E, Thompson M, Faraone SV, Asherson P, Johansson L (2006) The analysis of 51 genes in DSM-IV combined type attention deficit hyperactivity disorder: association signals in DRD4, DAT1 and 16 other genes. *Mol Psychiatry* 11:934–953
27. Wechsler D (1991) Wechsler Intelligence Scale for Children. The Psychological Corporation, London
28. Schallberger U (2005) Welches sind die nach statistischen Kriterien besten Kurzformen des HAWIK-III? (which are the best short forms of the WISC-III based on statistical criteria?). Research Report, University of Zurich
29. Sharpe D, Rossiter L (2002) Siblings of children with a chronic illness: a meta-analysis. *J Pediatr Psychol* 27:699–710