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A study to compare temperament in children with attention deficit hyperactivity disorder (ADHD) and their healthy siblings and association between ADHD symptoms and temperament dimensions in healthy siblings

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Abstract
Aim: This study aims to compare temperament in children with attention deficit hyperactivity disorder (ADHD) and their healthy siblings and to assess the association between ADHD symptoms and temperament dimensions in healthy siblings.
Material and Methods: A hundred and twenty children (60 children with ADHD and 60 siblings-one for each child with ADHD) were assessed retrospectively on temperament measurement schedule (TMS) and conners parent rating scale revised: short form (CPRS-R: S).Siblings of children with ADHD scored significantly lower than the probands on all subscales of CPRS-R: S.
Results: Mean age of the patients were 11.4 and mean education years in patients was 5.1. Majority of them were males (63.3%) and females were (36.7%) respectively. Persistence as a temperamental trait emerged to have a significant negative correlation with all the four subscales (oppositional, inattention, hyperactivity, ADHD index) of CPRS-R: S in siblings of children with ADHD.
Conclusion: Higher persistence in siblings appears to offer protection to these at-risk individuals who do not have ADHD; favoring the dual pathway model of ADHD.

Keywords: attention deficit hyperactivity disorder, siblings, temperament

Introduction

For many years, Attention-Deficit/Hyperactivity Disorder (ADHD) was believed to equally affect children of all social classes; that is, risk was not believed to vary by socioeconomic status (SES) [1-2]. This perception has persisted despite strong evidence that ADHD is more common among the poor, which has been documented in clinical samples [3-4], population-based epidemiologic studies [5-6], and large national surveys like the National Health Interview Study (CDC/NCHS, 2007), and the National Survey of Children's Health [7].

Given the strength of the evidence, it is puzzling that controversy exists; but two factors may play a role. First, low-income children without health care coverage are less likely to be treated for ADHD [8], which skews the pool of clinically identified patients toward more affluent children. The second factor is historical; the authors of two influential population-based studies of ADHD minimized SES in the etiology of ADHD when interpreting their results [9].

Temperamental characteristics of high negative reactivity, activity, novelty-seeking, impulsivity, low agreeableness/ hostility, low self-directedness, task persistence, low attentional focusing, inhibitory control, low conscientiousness/effortful control are associated with AD [10, 10-12, 14] and studied in clinical samples of children, [13-15] community samples [16] in whom effortful control is considered to mediate some of the executive functioning deficits. Despite this overwhelming evidence of association of temperamental characteristics with ADHD, coupled with their moderate heritability and their role as putative risk factors, there is scarce and inconclusive research regarding temperamental characteristics in first-degree relatives of children with ADHD [17-20].

Hence, we aim to compare temperament in children with attention deficit hyperactivity disorder (ADHD) and their healthy siblings and to assess the association between ADHD symptoms and temperament dimensions in healthy siblings.

Material and Methods

The study had a cross-sectional design and was carried out in the Department of Psychiatry MGM Medical College, Kishanganj, Bihar, India from March 2010 to Jan 2012

Ethical clearance was obtained from the Institute Ethics Committee and strict data confidentiality was maintained. Since, it was an exploratory study and no previous studies had examined child-sibling pairs, thus sample size was mainly drawn from clinical studies in children with ADHD alone and study sample of 60 children with ADHD and 60 healthy siblings (at least one for each child with ADHD) was drawn by non-probabilistic (convenience) sampling method. Child-healthy sibling pairs were recruited for the study.

Children of either gender aged 4–14 years diagnosed clinically with ADHD or hyperkinetic disorder according to DSM IV [18] or International Classification of Diseases 10 [19] and confirmed using the Mini International Neuropsychiatric Interview for children and adolescents (MINI KID) [20] and having a healthy sibling aged 4–14 years of either gender were included in the study. Healthy sibling was defined as a sibling of a child diagnosed with ADHD who had never been diagnosed with any psychiatric illness, was never referred by any teacher, medical or nonmedical agency, and had never sought psychiatric evaluation. The age range of 4–14 years

was considered to maintain homogeneity in the two groups of the study sample and also because temperament assessment in the index study was done till the 3rd year of age for both child-sibling pair (essentially to ensure that the same symptoms may not contribute toward measuring temperamental trait and symptom severity of ADHD). In addition, earlier clinic-based studies had included a broader age range including adolescents aged 14–18 and young adults. However, this was considered a limitation, as symptom profile in older adolescents and young adults may be different from those in the children and younger adolescents and therefore we restricted our study to children aged 4–14 years. The status was further confirmed by using the childhood psychopathology measurement schedule (CPMS) [21]. Those siblings with a CPMS score of >10, were assessed on MINI-KID and none were found to have any diagnosable psychiatric disorder. Those children with ADHD who had moderate to severe intellectual disability, autism spectrum disorder, epilepsy, or any other neurological disorder; did not have a sibling or did not provide assent/consent for the study were excluded. In case of siblings, those with any known psychiatric disorder, intellectual disability, epilepsy, or chronic physical disorder were excluded from the study. Written informed consent from parents and assent from all participants was obtained prior to recruitment into the study.

Temperament measurement schedule (TMS) which is an Indian adaptation of Thomas and Chess temperament questionnaire [22] was used for assessment of temperament till the 3rd year of age for both child-sibling pair. It is a bilingual parent interview schedule measuring nine temperamental traits, namely - Approach-withdrawal, adaptability, threshold of responsiveness, quality of mood, persistence, activity level, distractibility, and rhythmicity. Each temperamental trait is assessed based on 4 or 5 questions, scored from 1 to 5 (1-absence and 5-maximum level of manifestation). Five factors are derived from the nine traits, namely, Sociability (approach-withdrawal, adaptability, threshold of responsiveness), Emotionality (mood, persistence), Energy (activity level, intensity of reaction), distractibility and rhythmicity. TMS has been used in Indian children [23] and is found to be culturally valid.

Conners' parent rating scale-revised: Short form (CPRS-R: S) [24] is a 27-item 4-point Likert-type scale that yields 4 mutually exclusive scale scores defined by factor analysis: The cognitive problems/inattention scale (6 items), the hyperactivity scale (6 items), oppositional scale (3 items), and the ADHD index (12 items). It is a diagnostic instrument as well as used to assess the severity of ADHD symptoms. CPRS-R: S is also used as a screening tool and ADHD index subscale identifies children at high risk of ADHD. It has good reliability and validity with alphas between 0.86 and 0.94; and 6-week test-retest correlations between 0.72 and 0.85. [25]

Children with ADHD fulfilled the inclusion and exclusion criteria and their parents were approached for recruitment in the study. Those providing written informed consent were included in the study. Socio-demographic and clinical details were recorded in structured formats. Intelligence quotient (IQ) of probands was extracted from routine clinical records while IQ testing using standard progressive matrices/CPM [26] was carried out for all sibling participants. Parents were interviewed using the TMS for retrospective assessment of temperament and rating on CPRS-R: S for children with ADHD and their siblings.

Statistical analysis

The IBM SPSS Statistics for Windows, Version 20.0. Armonk, NY, USA: IBM Corp. was used for statistical analysis. Means and standard deviation (SD) were calculated for continuous variables and frequency and percentages for discontinuous variables. Student's t-test was computed to compare CPRS-R: S and TMS scores of probands and their healthy siblings. Analysis of covariance (ANCOVA) was used to control for gender while comparing the TMS and CPRS-R: S scores. Correlation matrix and analysis were carried out to assess the correlation of TMS with CPRS-R: S scores of siblings. Linear regression analysis by stepwise method was conducted to measure for the variance in ADHD subscales scores for siblings as assessed by CPRS-R: S explained by temperament dimensions as measured by TMS in them, depending on the results of correlation analysis.

Results

A total of 160 children clinically diagnosed with ADHD were screened for recruitment in the study. 90 patient-sibling pairs fulfilled the inclusion/exclusion criteria. The final sample with complete assessments included 60 patient-sibling pairs. Mean age of the patients were 11.4 and mean education years in patients was 5.1. Majority of them were males (63.3%) and females were (36.7%) respectively. [Table 1] Siblings of children with ADHD scored significantly lower than the probands on all subscales of CPRS-R: S, i.e., oppositional ($P < 0.001$), inattention ($P < 0.001$), hyperactivity ($P < 0.001$) and ADHD index ($P < 0.001$). ANCOVA showed that these differences remained significant even after controlling for gender. [Table 2]

Persistence as a temperamental trait emerged to have a significant negative correlation with all the four subscales (oppositional, inattention, hyperactivity, ADHD index) of CPRS-R: S in siblings of children with ADHD as shown in Table 3. Furthermore, distractibility had a significant positive correlation with the inattention subscale of CPRS-R: S. No other significant correlation was found between temperament dimensions and ADHD subscales.

To explain the variance in ADHD symptoms due to temperament, stepwise linear regression analysis was computed with ADHD subscales of CPRS-R: S as dependent variables and temperamental dimension of persistence as independent variable. The adjusted R square (here indicated as r^2) indicates the percentage of the variance of all explained by a variable or a set of variables. The results of regression analysis are shown in Table 4. Persistence explained 8.3% to22.4% of the variance of all subscales of CPRS-R: S. Persistence and distractibility together explained 24.7% of Inattention scores.

Table 1: Comparison of Sociodemographic profile of patients with attention deficit hyperactivity disorder and their siblings

Variables	Mean±SD Frequency [%]		P value Chi square
	Patient [n=60]	Sibling [n=60]	
Age (years)	11.4 (3.2)	10.8 (2.4)	1.9 (0.02)
Education (years)	5.1 (2.8)	4.3 (3.4)	7.8 (0.76)
Gender			
Male	38 (63.3%)	28 (46.6%)	$\chi^2 = 0.364$ (<0.05)
Female	22 (36.7%)	32 (53.7%)	

Table 2: Comparison of Conners Parent Rating Scale scores and temperament of children with attention deficit hyperactivity disorder with their siblings

Variables	Mean±SD		Mean Difference	P value
	Patient [n=60]	Sibling [n=60]		
CPRS-R: S				
Oppositional	8.1 (4.3)	2.5 (1.7)	5.6 (0.7)	< 0.001
Inattention	13.4 (3.6)	3.6 (1.6)	9.8 (0.8)	< 0.001

Table 3: Correlation of temperamental dimensions with Conners Parent Rating Scale domain scores of siblings

Temperamental Dimensions:	ADHD subscales			
	Oppositional	Inattention	Hyperactivity	ADHD index
Approach – Withdrawal	0.372 (0.226)	-0.194 (0.382)	0.064	0.78
Adaptability	0.483 (0.552)	-0.483 (0.261)	0.052	0.563
Threshold of responsiveness	-0.448 (0.162)	0.0338 (0.008)	0.631	0.462
Mood	-0.212 (0.432)	0.114 (0.410)	0.563	0.217

Table 4: Regression analysis: Variance of Conners Parent Rating Scale-Revised: Short form subscales explained by temperament measurement schedule dimensions

Dependent Variable (CPRS-R:S)	Predictor Variable	Subdural Variable
Oppositional	Persistence	-0.036
Inattention	Persistence	-0.042
Hyperactivity	Persistence	-0.063
	Divers ability	-0.079
	Persistence	-0.083

Discussion

For many years, ADHD was considered an exception because many mental health researchers believed that ADHD equally impacted rich and poor. Our data supports the hypothesis that ADHD is inversely related to socioeconomic status. Understanding this relationship is of considerable public health importance because ADHD is a common developmental disorder of childhood that is often accompanied by psychiatric comorbidity and substantial impairment in day-to-day functioning [7]. Interestingly, the healthy siblings in our study had only slightly less distractibility; this difference being non-significant. Persistence refers to continuation of an activity in the face of obstacles to the maintenance of the activity direction, [27] and Distractibility is the effectiveness of extraneous environmental stimuli in interfering with or in altering the direction of ongoing behavior [27]. Hence, our healthy siblings had almost same degree of Distractibility, but with higher Persistence, they could again come back to the task and maintain the activity direction. Persistence is similar to Effortful control in Rothbart and Bates model, that describes the ability to suppress a dominant or automatic response, persist with the non dominant response, pay attention, detect errors and plan for future. Both persistence and effortful control reflect the regulatory aspect of temperament, as opposed to reactive aspects [28, 29]. Epigenetic research on how a person's genes and their social environment interact to shape their risk of psychopathology [30] suggests a compelling avenue of research for understanding the etiology of ADHD and its comorbidity. Goldsmith *et al*., [31] reported biometric model fitting on longitudinal twin data. They found that both genetic and environmental sources of variance in effortful control accounted for variance in later ADHD symptoms. Furthermore, all the genetic variance in later ADHD symptoms was in common with the genetic variance from earlier effortful control. Our finding in healthy siblings of clinically diagnosed cases is in keeping with the observation of Goldsmith [31]. The authors proposed that temperament presents as a liability to childhood psychopathology.

Conclusion

Higher persistence in siblings appears to offer protection to these at-risk individuals who do not have ADHD; favoring the dual pathway model of ADHD.

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