ORIGINAL ARTICLE

Attention deficit hyperactivity disorder and intellectual giftedness: a study of symptom frequency and minor physical anomalies

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Objective: To evaluate the presence of symptoms of attention deficit and hyperactivity disorder (ADHD) in intellectually gifted adults and children.

Methods: Two cross-sectional studies were performed in children and adults whose intelligence quotient (IQ) had been previously evaluated using Raven's Progressive Matrices (RPM) test. Seventy-seven adults displaying IQ scores above the 98th percentile were assessed using the Adult Self-Report Scale (ASRS-18) for signs of ADHD and a modified Waldrop scale for minor physical anomalies (MPAs). Thirty-nine children (grades 1-5) exhibiting IQ scores above the 99th percentile, as well as an equally matched control group, were assessed for ADHD by teachers using the Swanson, Nolan and Pelham IV Rating Scale (SNAP-IV) as used in the NIMH Collaborative Multisite Multimodal Treatment Study of Children with Attention-Deficit/Hyperactivity Disorder (MTA-SNAP-IV).

Results: In gifted adults, the frequency of ADHD-positive cases was 37.8%, and the total MPA score was significantly associated with ADHD (p < 0.001). In children, the ADHD-positive case frequency was 15.38% in the gifted group and 7.69% in the control group (odds ratio [OR] = 2.18, p = 0.288). **Conclusions:** The high frequency of ADHD symptoms observed, both in gifted adults and in gifted (and non-gifted) children, further supports the validity of this diagnosis in this population. Furthermore, the significant association between MPAs and ADHD suggests that a neurodevelopmental condition underlies these symptoms.

Keywords: ADHD; giftedness; minor physical anomalies; twice exceptional

Introduction

There has been much controversy as to whether attention deficit and hyperactivity disorder (ADHD) symptoms could be present in highly intelligent individuals and, if present, whether those symptoms are indeed ADHD, some other condition, or overlapping ADHD-like characteristics of giftedness itself. The relative scarcity of scientific data in the field so far has fueled this debate, and mental health professionals and educators frequently disagree on the subject. Within this context, the present study sought to conduct an assessment of ADHD symptoms in adults and children whose intelligence quotient (IQ) is at the upper end. This investigation was conducted in a rather large sample, in an attempt to ensure adequate representation of gifted individuals, who were recruited using a convenience strategy. As a secondary objective, the association of minor physical anomalies (MPAs) and ADHD was also evaluated in adults in order to compare ADHD with other neurodevelopmental conditions and explore its clinical utility. The causes of MPAs include both genetic and fetal environmental factors, such as perinatal asphyxia and other insults to normal pregnancy that affect the ability of the genome to channel development towards a normal phenotype. This concept is known as Waddington's channeling notion, and is the foundation of developmental instability theory.¹

Intellectual giftedness has always been a fascinating theme, even though intelligence itself has always been rather difficult to define and measure. Several authors in the field have argued that giftedness as well as intelligence must be evaluated by multidimensional approaches. However, in practical terms, the IQ score is widely used to evaluate intelligence, as it correlates positively with other cognitive skills, especially those related to reasoning and spatial abilities.² There are two apparently conflicting ideas regarding the intellectually gifted: the first is that high IQ is a protective factor for psychopathological phenomena ("the cognitive reserve" concept); the second arises from the anecdotal idea that "geniuses" are eccentric, distracted, careless, and lack common sense. It is also a common observation that a number of intelligent individuals fail to perform accordingly in their academic, professional, and interpersonal areas, 3,4 and, recently, Duckworth et al. proposed that self-discipline is a better predictor of performance than

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Submitted Jul 06 2014, accepted Dec 30 2014.

IQ.⁵ Gifted individuals with disabilities have been lately labeled as "twice-exceptionals," even though not much is known about how intellectual giftedness might relate to psychopathologies, which may play a negative role in the performance of these individuals. A few disorders are already known among "twice-exceptionals," such as Asperger's syndrome. Some authors propose that a degree of autism (or "savantism") might enable such high abilities in the gifted. Recent studies have also mentioned bipolar mood disorder and higher rates of illegal substance use among high-IQ individuals. A lesser known theory that described the combination of mental illness, intellectual giftedness, and MPAs was published in the late 1960s by Salles, who compiled a large case series and developed a model for perinatal asphyxia to explain the co-occurrence of neurodevelopmental diseases and intellectual giftedness. 12

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According to recent studies, 13,14 ADHD affects about 5.3% of children and adolescents, and about 2.9% of the adult population. A meta-analysis concluded that patients with ADHD had lower cognitive ability than control individuals¹⁵; however, this finding was later shown to be due to a small subset of individuals (females with birth complications) with IQ scores 15 points below those of the overall control group. 16 Other studies suggest that IQ is distributed normally within the ADHD population, and that it can be a predictor of better performance and later medical evaluation. 17-19 A recent neuroimaging study 20 demonstrated that the co-occurrence of ADHD and high IQ has a different neuroanatomical behavior then the other types (ADHD with normal IQ and ADHD with low IQ).2 While IQ correlated positively with gray matter volume (r = 0.31) in controls, in the ADHD/high-IQ group, this correlation was negative (r = -0.25). The authors of this study suggest the existence of multiple phenotypes for ADHD using IQ, and a particular one for the co-occurrence of ADHD and high IQ. Some studies have found that high IQ can be a predictor of medication response,²¹ further suggesting that ADHD in the high-IQ population might even have a different pathophysiology than ADHD in individuals with normal IQ.

Despite this evidence, several psychologists and educators deny the validity of an ADHD diagnosis in intellectually gifted subjects even when symptoms are present, usually referring to Dabrowski's model of overexcitabilities.²² Currently, the Brazilian Council for Giftedness (CONBRASD) states on its web page that "The common symptoms of ADHD (inattention, impulsivity and hyperactivity) often lead to misdiagnosis of a child with high abilities/giftedness as hyperactive" (http://conbrasd. org/wp/?page_id=4188). This resistance notwithstanding, support is building for the validity of ADHD diagnosis in gifted individuals. The co-occurrence of high IQ and ADHD in children 18,23,24 and adults 25,26 has been documented in several studies from ADHD centers. A Brazilian case series observed the co-occurrence of ADHD and high IQ in a referred population, noting other frequent conditions in this group,²⁷ and, according to Brown,²⁵ adults with high IQ and ADHD tend to experience greater deficits in function than the general population. Finally, none of the studies mentioned so far have focused on participants with IQ > 132 (> 98th percentile).

Methods

Ethics

All experimental protocols and the design of the study were approved by the Ethics Committee of Universidade de São Paulo (USP), SP, Brazil. The Brazilian branch of the Mensa society (Mensa Brazil), from where the adult sample was drawn, gave their consent for the study and all included subjects provided individual written informed consent. Parental permission was obtained for the participation of children in the study, and written informed consent was provided by the participants' teachers.

Sample

The investigation consisted of two separate crosssectional studies of adults and children. Both were convenience samples, as both had been previously tested with Raven's IQ test (Advanced [APM] version for adults and Colored [CPM] for children).

Inclusion criteria for adults

Individuals were recruited among 158 active members of Mensa (a high IQ society for individuals with IQ scores at or above the 98th percentile, http://www.mensa.org/) residing in the state of São Paulo during the second semester of 2012. Of these, 77 (49%) were included in the study, 41 (26%) did not respond to the invitation, 26 (16%) refused to participate, 9 (6%) met the sole exclusion criterion (self-declared diagnosis of a psychiatric condition other than ADHD), and 5 (3%) had invalid contact information. Of the 77 included participants, 78% were male and 82% chose to have their data (except for the Adult Self-Report Scale [ASRS]) collected with investigator assistance via a personal interview or by teleconference. Participants in the unassisted group completed online versions of the ASRS and of the modified Waldrop scale using a web-based form containing pictures and descriptions. Comparisons between the included group the group of losses and we observed no significant difference in gender (p = 0.47) or age (p = 0.36). No control group was used due to logistics limitations.

Inclusion criteria for children

All student with IQ scores > 99th attending grades 1-5 of primary education at the Colégio Objetivo program for gifted students (http://www.objetivo.br/poit/) as of December 2012 were included in the study, for a total of 39 individuals (70% males). The control group consisted of 39 other students, matched for sex and age, randomly selected from the same classes but with IQ scores \leq 90th percentile. There were no exclusion criteria.

Measures

ADHD in adults

To assess ADHD in adults, a Brazilian version²⁸ of the World Health Organization Adult ADHD ASRS²⁹ 1.1 scale

was used. The ASRS scale is a widely standardized tool that has been used in large population studies in the U.S.³⁰ and Brazil.³¹ Version 1.1 is composed of 18 questions, nine referring to inattention and the reminder to hyperactivity-impulsivity (HI). We used three criteria to determine ADHD cases (research screening diagnosis) and calculate the positive case frequency: first, according to Kessler et al., 29 scores above 24 (range, 0-72) are indicative of ADHD, but this symptomatic threshold has not been widely validated.²⁸ Second, the shorter (sixitem) version of this scale is easily obtained by its deconstruction and has another case-defining criterion.30 The final criteria were developed by the authors to increase conservativeness and include some of the DSM-IV criteria for ADHD that are overlooked by the questionnaire: a) symptom onset before 7 years of age: b) universality of symptoms, such as manifestation in at least two different environments: c) clinically significant functional impairment; and d) clinical decision that symptoms are not attributed to other disorders, e) especially mood and anxiety disorders. Screening was conducted by a clinically experienced, board-certified psychiatrist.

ADHD in children

A translated version³² of the NIMH Collaborative Multisite Multimodal Treatment Study of Children With Attention-Deficit/Hyperactivity Disorder (MTA) - Swanson, Nolan, and Pelham IV Rating Scale (MTA-SNAP-IV), a tool widely used for child ADHD research,³³ was applied by teachers to the sample of grade 1-5 students.

The scale consists of 26 questions scored according to symptom frequency. The first nine items are related to inattention symptoms, the following nine items assess HI symptoms, and the last eight items evaluate symptoms of oppositional defiant disorder. The scale's author has defined weighted sub-scores for inattention and HI, as well as a combined score; because the sub-scores are weighted, each ranges from 0 to 3. A cutoff score of the top 5% is defined as significant for ADHD. According to the author, this would correspond to a teacher-assigned rating of 2.56 for inattention, 1.78 for HI, and 2 for the combined score (also known as the SNAP-IV criterion).34 Slightly lower scores of 1.2 for HI and 1.8 for inattention were recently proposed by a large study (also known as the Bussing criterion).33 Finally, we added a DSM-IVbased criterion, which defines ADHD positivity as presence of at least six symptoms from the ADHD axis (items 1-18) rated as frequent or very frequent. This is similar to the criterion used in the adults, but with no evaluation of the other DSM-IV criteria. The complete set of criteria is described in Table 1.

Minor physical anomalies (MPAs)

MPAs were evaluated in adults using a modified Waldrop scale,³⁵ administered by a researcher who, at the time, was blind to ASRS results. Participants who were not physically examined (unassisted group) were allowed to self-report MPAs in graphic form. The Waldrop scale has been used in

mental illness research for 40 years and has become a *de facto* standard, however it has many weaknesses because it is more of a checklist with loose parameters than a real scale and no data on general population or different ethnic groups exists. It has been employed in studies of autism,³⁶ schizophrenia,³⁷ and hyperactivity,³⁸ among others.³⁹ Based on the literature, six other physical signs were added to the scale¹²: 1) joint hypermobility; 2) "bayonet finger"; 3) genu valgum; 4) flat foot; 5) 4D:2D finger ratio; and 6) strabismus divergens. Ismail's modified Waldrop scale³⁵ consists of the following items:

- Head: a) abnormal head circumference; b) abnormal hair whorls; c) fused eyebrows; d) frontal bossing; e) micrognathia; f) anteverted nostrils; g) eyes: telecanthus, epicanthus, ptosis, and heterochromia; h) Ears: adherent ear lobes, malformed ears, asymmetric ears, low-seated ears, and ear lobe skin tag; i) mouth: high/steepled palate, furrowed tongue, tongue with smooth-rough spots, and thin upper lip:
- Hands: a) clinodactyly; b) abnormal palmar crease;
 b) hyperconvex nails; d) arachnodactyly; e) hypoplastic nails;
- 3. Feet: a) large gap between 1st and 2nd toes; b) partial syndactyly; c) retarded toe (4th or 5th); d) deep crease between the 1st and 2nd toes; e) hyperconvex nails; f) overlapping of the 4th and 5th toes.

Statistical analysis

Data were stored and processed using the GNU R statistical system (http://www.r-project.org) and the epicalc epidemiology library (http://cran.r-project.org/web/packages/epicalc).

Adults

The different data collection methods were divided into subgroups and an analysis of variance (one-way ANOVA) was performed to assess possible distortions in MPA data from the unassisted group. The distribution of MPAs was then tested with the Shapiro-Wilk normality test, and homogeneity of variances was tested with the *F*-test. To assess differences in mean, we used the non-parametric Mann-Whitney *U* test for non-normal distributions. Risk analysis was done using the Newcombe-Wilson method and Fisher's chi-square test. An odds ratio (OR) analysis and Fisher's chi-square test were then performed to verify each individual MPA and ADHD. Finally, results were evaluated by the Mantel-Haenszel method to check for gender influence.

Children

Another Newcombe-Wilson risk analysis method was used to compare the test group (gifted individuals, IQ > 99th percentile) with the control group (sex- and agematched individuals with IQ < 90th percentile). A similar test was used in an IQ percentile stratified analysis, the last stratum being comprised solely of the gifted group. We also evaluated these results using the Mantel-Haenszel method to check for gender influence.

Table 1 Case definition criteria for ADHD

Criterion	Description
Adults ASRS-18 ASRS-6 DSM-IV based	Score ≥ 24 Score ≥ 4 (obtained by deconstruction of ASRS-18) Six or more symptoms of inattention or hyperactivity/ impulsivity scored at least as often + onset before age 7 years
Children SNAP-IV Bussing DSM-IV based	Score ≥ 2.56 for inattention or ≥ 1.78 for HI or combined score ≥ 2 Score ≥ 1.8 for inattention or ≥ 1.2 for HI At least six ADHD symptoms rated as frequent or very frequent

ADHD = attention deficit and hyperactivity disorder; ASRS = Adult Self Report Scale; HI = hyperactivity-impulsivity; SNAP-IV = Swanson, Nolan, and Pelham IV Rating Scale.

Results

Adults

We obtained very different frequencies of ADHD positivity depending on the criterion used (Table 1), as follows: ASRS-18, 82%; ASRS-6, 51%; DSM-IV based, 38%.

The mean ASRS-18 score was 38.14 ± 9.27 and the mean number of MPAs was 5.6 ± 4.67 . Table 2 describes the age, ASRS-18 and ASRS-6 scores, sub-scores, and MPAs for the adult subjects assessed.

We then performed ANOVA to compare total MPA scores between the assisted and unassisted groups, which indicated that the latter had a significantly smaller ($\Pr[>F]=0.003$) number of MPAs; however, a similar analysis of ASRS score did not demonstrate significant differences ($\Pr[>F]=0.334$). We also found no significant difference between the means and distributions of attentional and HI sub-symptom scores.

DSM-IV-based criteria were used to define ADHD cases for MPA analysis. Unadjusted MPA results were analyzed both using the total number of anomalies and by each anomaly individually. The ADHD-positive group had a mean MPA score of 8.31 ± 4.61 , vs. only 3.96 ± 3.91 in the ADHD-negative group. MPAs were not normally distributed per the Shapiro-Wilk test (W = 0.90; p < 0.001), and the *F*-test demonstrated non-homogeneity of variances of MPA in the ADHD-positive (greater variance) and ADHD-negative groups (F=0.72; p = 0.32). Finally, the Mann-Whitney *U*-test indicated a true location shift greater than zero toward the ADHD-positive group (W = 1057; p < 0.0001). Individual anomalies presenting a significant association with ADHD (p < 0.05) are shown in Table 3.

A gender-stratified Mantel-Haenszel analysis of every anomaly listed in Table 3 indicated greater significance and higher ORs for males, except for high/steepled palate.

Children

As in adults, the frequency of ADHD positivity varied depending on the criteria used do define it. Risk and OR analysis demonstrated no statistically significant differences in the frequency of ADHD positivity between the gifted group and the control group in any criterion, as shown in Table 4.

As in adults, a gender-stratified Mantel-Haenszel analysis revealed no significant gender influence (p = 0.39) in the frequency of ADHD positivity. Using the SNAP-IV criterion to define ADHD cases, we conducted a risk analysis of ADHD stratified by IQ percentile in the whole sample (controls and gifted subjects) and obtained χ^2 = 4.91 and p = 0.16 (Table 5).

Discussion

A high frequency of ADHD symptoms was observed in all groups, regardless of the criteria used to define cases. The

Table 2 Age, ASRS-18 and ASRS-6 scores, and MPAs in gifted adult subjects

Variable	Adult subjects (n=77)	ADHD+	ADHD-
Age	36.92±9.27 (20-64)	34.41±8.62 (20-58)	38.44±9.27 (27-62)
ASRS-18	38.14±13.15 (12-64)	47.45±6.62 (37-64)	32.52±12.85 (12-62)
Inattention sub-score	19.36±7.50 (6-33)	24.76±4.56 (17-33)	16.10±7.05 (6-32)
Hyperactivity/impulsivity sub-score	18.78±7.55 (4-32)	22.69±5.75 (6-32)	16.42±7.56 (4-31)
MPA	5.60±4.67 (0-15)	8.31±4.61 (0-15)	3.96±3.91 (0-14)
ASRS-6	3.40±1.56 (0-6)	4.76±0.99 (3-6)	2.58±1.54 (0-6)

Data presented as mean ± standard deviation (range).

ADHD = attention deficit and hyperactivity disorder; ASRS = Adult Self-Report Scale; MPA = minor physical anomalies.

Table 3	Minor	physical	anomalies	significantly	associated with	ADHD

Sign	OR	95%CI	χ^2	p-value
Abnormal hair whorls	8.03	2.19-32.53	13.94	0
Clinodactyly	4.61	1.55-13.98	9.75	0.002
Strabismus divergens	4.41	1.48-13.26	9.20	0.002
High/steepled palate	4.25	1.42-12.81	8.7	0.003
Large gap between 1st and 2nd toes	4.07	1.33-12.62	7.87	0.005
Bayonet finger	3.53	1.08-11.77	5.74	0.017
Thin upper lip	3.06	0.96-9.83	4.67	0.031
4D:2D finger ratio	2.62	0.89-8.18	3.74	0.053

95%CI = 95% confidence interval; ADHD = attention deficit and hyperactivity disorder; OR = odds ratio.

Table 4 Odds ratio analysis and frequency of ADHD-positive cases in children by case definition criteria and group

Criterion	ADHD+ in gifted group (%)	ADHD+ in control group (%)	OR	95%CI	χ²	p-value
SNAP-IV	15.38	7.69	2.18	0.42-14.43	1.13	0.288
Bussing	17.95	12.82	1.49	0.36-6.56	0.39	0.53
DSM-IV-based	17.95	25.64	0.63	0.18-2.14	0.68	0.411

95%CI = 95% confidence interval; ADHD = attention deficit and hyperactivity disorder; OR = odds ratio; SNAP-IV = Swanson, Nolan, and Pelham IV Rating Scale.

Table 5 Risk analysis of ADHD in all children (gifted subjects and controls) stratified by IQ percentile

IQ percentile	Absolute risk	Risk ratio	95%CI	ADHD+	ADHD-
20th-40th	0.29	1	Null	2	5
40th-60th	0	0	∞-0	0	10
60th-80th	0.50	0.16	4.14-0.01	1	21
80th-100th	0.15	0.54	28.37-0.01	6	33

95%CI = 95% confidence interval; ADHD = attention deficit and hyperactivity disorder; IQ = intelligence quotient. The last row is composed solely of the test group (> 99th IQ percentile).

37.8% frequency of ADHD in gifted adults (obtained using the DSM-IV-based criterion) is much higher than that expected for the Brazilian population, which has been estimated by our group as 5.8%, using the ASRS-6 in a nationally representative sample of adults. We acknowledge that this comparison lacks methodological rigor; nonetheless, we obtained a 51% frequency when using the same criterion (ASRS-6) as in the national study mentioned. The high frequency of ADHD cases among gifted children is also worthy of note, even though no statistically significant difference was found between the gifted and control groups (p = 0.28).

Some caveats and methodological limitations of this work must be mentioned. The participants were recruited via a convenience sampling strategy, which could be a source of selection bias; we lacked a control group for the adult subsample and had limited access to the children to further evaluate MPAs and other DSM-IV criteria. The same clinically experienced psychiatrist evaluated all adult participants (increasing reliability in this case), but no structured psychiatric diagnostic tool (e.g., the Composite International Diagnostic Interview [CIDI]) was employed, and the authors empirically observed that other disorders (especially autism spectrum disorders) might be highly frequent among participants, despite no formal prior diagnosis. Our methodology allowed only a screening diagnosis for research purposes. In addition, we acknowledge that selection bias exists relative to our responsiveness in the adult study; however, the direction of this bias is impossible to determine. Participants with an interest in ADHD tend to easily accept inclusion; on the other hand, difficulties inherent to ADHD should make engagement in research protocols more difficult. We are also unaware of the reasons that make an individual eligible to seek membership and remain active in Mensa, which may also be a potential source of bias. Thus, our findings should be extrapolated to the gifted adult population with extreme caution.

Regarding potential biases in the pediatric subsample, we considered that, given that some believe that gifted children may display overlap in behaviors common to ADHD, it is possible that ADHD parents sought to enroll their children in this particular school because of its special program for gifted students. This potential bias

would explain the large number of children diagnosed with ADHD in the control group. Data from an age-, grade-, and sex-matched control group composed of students from a similar private teaching institution that does not run special programs for the gifted would clarify this issue.

Additionally, psychometric research tools are inherently inaccurate (i.e., measurement error), and the ASRS scale is a self-assessment questionnaire; hence, all ADHD symptom data we obtained was restricted to self-reporting (adult sample) or teacher reports (children). Given the restrictions mentioned above, we sought to use the most conservative criteria for ADHD case definition, preferably based on DSM-IV (although full DSM-IV criteria were never met). This corresponded to the DSM-IV-based criterion for the adults. However, the SNAP-IV criterion used for the children was most conservative. The stratified risk analysis shown in Table 5 also had limitations due to the small number of participants and uneven distribution of IQ percentiles; however, its findings do draw attention to the zero-risk stratum precisely at the middle (between the 40th and 60th percentile) and to increased risk toward the upper and lower strata.

A relationship between MPA and ADHD was found in this sample. Furthermore, of the 36 MPAs analyzed, eight had significant individual associations with ADHD. There are no reliable data on the frequency of these signs in the general population, nor any ethnic validation, which makes MPAs a marker with low specificity and unknown sensitivity. Therefore, their association with ADHD symptoms should be regarded primarily as an exploratory finding. Nevertheless, despite the limitations mentioned, these phenomena may have clinical potential, particularly those located in the hands and head (which are easily observed). In addition, our finding agrees with past clinical observations of associations between MPAs and several conditions, including hyperactivity itself.³⁸ The higher number of MPAs in gifted adults with ADHD suggests a pattern of developmental changes that may result from or be associated with minor cerebral alterations, consistent with the neurodevelopmental model of ADHD.

Our findings may suggest that ADHD symptoms are not only present but also highly frequent among the intellectually gifted. Despite the restrictions mentioned above, this should at least prompt further investigations on this topic. In addition, ADHD diagnoses should be considered in the gifted population, contrary to what has been proposed by some groups in the field. We also believe the results of our study will contribute to confirming the existence of ADHD symptoms among the highly gifted, to strengthening the validity of the ADHD diagnosis in high-IQ children²³ and adults,²⁶ and to demonstrating their associations with MPA and possible deviations from normal development. Finally, we hope our findings will add to and improve the literature on psychopathology at the upper end of intelligence, which has turned out to be more complex than previously thought.

Acknowledgements

This work was supported in part by the following Brazilian government agencies: Conselho Nacional de Desenvolvimento Científico e Tecnológico (CNPq), Coordenação de Aperfeiçoamento de Pessoal de Nível Superior (CAPES), and Fundação de Amparo à Pesquisa do Estado de São Paulo (FAPESP). The authors thank Cristiane Cruz and Douglas Honório (president and secretary of Mensa Brazil), Christina Cupertino, Magali Maldonado, and Malu Silva (Colégio Objetivo), and Clarissa Paim (PRODAH HCPA).

Disclosure

LAR has served on the speakers' bureau and/or acted as a consultant for Eli Lilly, Janssen-Cilag, Novartis, and Shire, in the last 3 years. He also received travel fees (airfare and hotel costs) from Novartis and Janssen-Cilag in 2010 for taking part in two child psychiatry meetings. The ADHD and Juvenile Bipolar Disorder Outpatient Programs chaired by him received unrestricted educational and research support from the following pharmaceutical companies in the last 3 years: Abbott, Eli Lilly, Janssen-Cilag, Novartis, and Shire. DM reports no conflicts of interest.

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