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Research Article

Neuropsychological Profiles and Behavioral Ratings in ADHD Overlap Only in the Dimension of Syndrome Severity

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Objectives. The aim of this study was to compare the cognitive neuropsychological and the behavioral rating profiles of attention deficit/hyperactivity disorder (ADHD). *Methods.* Forty-two children diagnosed with ADHD (M = 11.5 years, SD = 1.1) and 43 typically developing children (M = 11.2 years, SD = 1.7) participated. We measured symptom severity with behavioral rating scales, and we administered neuropsychological tasks to measure inhibitory performance, updating/working memory, and shifting ability. *Results.* On the basis of the three neuropsychological variables, the hierarchical cluster analytic method yielded a six-cluster structure. The clusters, according to the severity of the impairment, were labeled as follows: none or few symptoms, Moderate inhibition and mild shifting, moderate to severe shifting. There were no systematic differences in inattention and hyperactive-impulsive behavior across the clusters. The comorbid learning disorder appeared more likely only in severe neuropsychological forms of ADHD. *Conclusion.* In sum, our results suggest that behavioral ratings and neuropsychological profiles converge only in the dimension of symptom severity and that atypicalities in executive functions may manifest in nonspecific everyday problems.

1. Introduction

Attention deficit/hyperactivity disorder (ADHD) is one of the most frequent psychiatric disorders affecting approximately 5% of children [1, 2]. Despite its wide effects on social and academic achievements, the diagnosis has remained controversial [3]. The diagnosis of ADHD (DSM-5; [4]) is mostly based on clinical observations, parental (and rarely teacher) interviews, or rating scales, but the neuropsychological information is often missing from the protocol [5]. However, given the temporal and methodological instability (i.e., combining information from different sources) of the diagnosis [3], assessment cannot focus only on the behavioral ratings. Moreover, in order to develop a sufficient treatment plan, it is necessary to evaluate various impairments—including cognitive deficits in attention, executive functions (EF), and memory—affecting day-to-day functioning, and to

determine the presence of any deficiency in adaptive skills and key competences. To point at this shortcoming of the diagnostic protocol, the aim of this study was to compare the cognitive neuropsychological and the behavioral rating profiles of ADHD.

With respect to the neuropsychological impairments, children with ADHD show deviations in executive functions, including inhibitory control, delay aversion, and time estimation [6, 7]. Several studies proved the dysfunction of working memory (WM); however, this impairment is not specific to ADHD, and it can be observed in many other developmental psychiatric syndromes, such as autism spectrum disorder, conduct disorder, or oppositional defiant disorder [6, 8, 9]. EF assessments vary in a broad range; therefore, a selection of EF measures that is not theory-driven could increase the heterogeneity of neuropsychological findings in ADHD [9, 10]. Miyake et al. [11] used confirmatory factor analysis

(CFA) to understand the relationships among three types of executive functions: mental set shifting, inhibiting prepotent responses, and updating the contents of WM. The three functions were not only moderately correlated but also clearly separable. Further studies used this model in community based developmental and child clinical settings as well [12–14]. These studies used the three-factor model [11] with different EF tasks and corroborated the robustness of this model.

Though there is substantial research on this topic spanning a large period of time (with varying study designs), the relationship between symptoms of ADHD (inattentive, hyperactive-impulsive behavior) and EF is still unclear [9, 15]. Nigg et al. proposed an "executive deficit type" within the category of ADHD [16]. This suggestion was based on an estimation that only 35-50% of children with ADHD have inhibitory deficit. In line with the notion that ADHD can develop in multiple pathways [13, 17–19], a subtype with EF impairment as a potential endophenotype could lead to targeted etiological research and personalized treatment, as well. The study of Lambek et al. [20] investigated the cognitive and academic performance of children with ADHD with and without an executive function deficit (EFD). While the ADHD-EFD group was characterized by lower IQ and higher intraindividual response variability, children with ADHD without EFD showed more delay aversion but otherwise intact EF and IQ. The authors suggested [20] that an EFD subtype could represent different risk factors and different needs for educational and clinical care.

Another study used two-step cluster analysis to detect profiles of children with ADHD with distinguishable neuropsychological profiles [21]. They found a three-cluster structure, where the profiles represented children with poor inhibitory control, poor set shifting/speed, and intact task performance, respectively. Despite the importance of working memory in cognitive development [6, 8], they did not investigate WM. Participants from the poor set-shifting/speed cluster had more hyperactive-impulsive ADHD and ODD symptoms and lower IQ than children from the other clusters. Despite the fact that atypical inhibition is often described as a major cognitive characteristic of ADHD [6, 16, 22], the poor inhibitory control cluster did not show more risk for ADHD than the other two clusters [21]. At the same time, children with ADHD from the intact task performance cluster had more severe depression symptoms. The study targeted children with ADHD only; therefore, the cluster structure of EF in nonclinical children remained unfolded. However, other studies found mild or moderate EF impairments in nonreferred samples as well [13, 16].

Another cluster analytic study used a multimeasure, multi-informant approach based on ADHD rating scales with a preschooler community sample (hierarchical cluster analysis with Ward's method, [7]). They found four clusters, where the "high comorbidity risk" cluster showed the lowest inhibitory control performance and the "ADHD risk only" cluster showed the highest level of delay aversion. The two further clusters were characterized by few/none symptom and by sensorimotor deficits without EF impairments, respectively. Other studies could not present such clear difference between ADHD and typically developing (TD) groups (e.g., [9, 13, 16]). Sjöwall et al. [13] found that children with ADHD had weaker performance than nonclinical children in working memory, inhibition, shifting, and emotion recognition in general, accompanied with greater reaction time variability. There were no group differences in delay aversion and in recognition of disgust. However, only one-third of the clinical participants had impairments in executive functions according to the 90th percentile of the nonclinical group's neuropsychological performance which was used as a cutoff criterion for what was regarded as impaired. More importantly, 26% of the nonclinical group had at least one neuropsychological deficit.

Our aim was to investigate whether subgroups characterized by various EF impairments are identifiable in a mixed sample of TD children and children with ADHD. Moreover, we tested how these expected clusters (see the following) differ in ADHD symptoms and comorbid conduct disorder (CD) and learning disorder (LD) problems. Given the diversity of EF [11] and the multifactorial heterogeneity of ADHD [21], we assumed that there would be at least five different cognitive clusters in our sample, and each one would be characterized by different symptom dimensions according to the rating scale scores. Our expectations were based on the conceptually relevant combinations of the three EF factors [11] and the two dimensions of ADHD symptoms. We expected a group of children with atypical inhibition, shifting, and updating associated with inattentive and hyperactive-impulsive symptoms, one cluster with solely updating problems and characterized by inattentive behavior, one with impairment in inhibition and high hyperactiveimpulsive ratings, a group characterized by shifting problems related to both dimensions of ADHD symptoms, and a last group with normal neuropsychological profile and few or no behavioral problems.

2. Methods

2.1. Participants and Procedure. Eighty-five children were invited to participate in the present study. Clinical participants (38 boys, 4 girls, M = 11.5 years, SD = 1.1) were recruited from the Vadaskert Child Psychiatric Clinic where they were diagnosed with ADHD by a group consisting of a psychiatrist, a psychologist, a neuropsychologist, and an expert on special education. The diagnoses were based on the DSM-IV-TR [23]. All of the clinical participants met the criteria of ADHD-C in regard to their symptoms. The members of the typically developing group (36 boys, 7 girls, M = 11.2 years, SD = 1.7) were recruited from a primary and a high school in Budapest. The age range was from eight to fifteen years in both groups. Parents of all participants provided informed consent and the children made an oral agreement. The research was granted by the Medical Research Ethics Committee of Semmelweis University. Those children with ADHD, who strongly manifested comorbid disorders (autism spectrum disorder, obsessive-compulsive disorder, Tourette syndrome, or major depression) and/or low socioeconomic status, were excluded from the study. Children with learning disorder scored below appropriate age level of Hungarian Logopaedic Test Protocol [24], including tests of numerical cognition, communicative development, spontaneous speech, phonological awareness, and grammar and vocabulary. The children did not meet the criteria of specific language impairment, dyscalculia, or dyslexia. Participants of the TD group who possessed any psychiatric or neurological records were also excluded. An additional exclusion criterion was an estimated IQ below 80 (based on Raven Progressive Matrices [25]) in both groups. The two groups did not differ in gender $\chi^2(1) = .34$, P = .56. Members of the ADHD group were significantly older than those in the TD group, t(195) = -7.64, P < .001; by this reason we also tested the effect of age on cluster structure.

2.2. Measures

2.2.1. Questionnaires. The following questionnaires were administered: ADHD Rating Scale (ADHD-RS, [26]), the Children's Depression Inventory [27], the Yale Global Tic Severity Scale [28], and the Child Yale Brown Obsessive Compulsive Scale [29]. The children were examined in accordance with the MINI International Neuropsychiatric Interview for Children and Adolescents (MINI-KID, [30]) semistructured interview. The results of the questionnaires and interviews listed above are not presented in this paper, except for the ADHD Rating Scale.

2.2.2. Neuropsychological Measures

Golden Stroop Test. The Golden Stroop test [31] was administered in which participants were required to name as many items as they could in 45 seconds for each of the three cards (word, color, and color-word). The outcome variable used in the analyses was the interference score as an indicator of prepotent response inhibition or cognitive conflict.

Digit Span. To measure the updating factor, the digit span backward task of the Wechsler Intelligence Scale of Children (WISC-III, [32]) was administered, which is the most widely used test for working memory [33].

Wisconsin Card Sorting Test (WCST). The original 128-card version of the WCST was administered [34]. In this test participants saw four stimuli cards and two packs (2×64) of response cards. The stimuli cards differed in color, number, and form. Participants were asked to match the response cards to the stimuli cards with consideration of the feedback (correct/incorrect) given by the experimenter. The matching rules changed after every ten correct answers to which participants were blind. For analyses, we used the number of perseverative errors which is an indicator of problems in mental set shifting [11].

2.3. Statistical Methods. For identifying subgroups with distinct patterns of the three main executive factors, an agglomerative hierarchical cluster analysis was conducted, for which the interference score of the Golden Stroop test and the digit span backward score and the number of perseverative errors from the Wisconsin Card Sorting Test were used as clustering variables. We applied squared Euclidean distance as the similarity measure and Ward's method as the type of cluster fusion, which was found to be more accurate and effective than solutions yielded by other techniques [35]. We did not use usual standardization methods on the three clustering variables but quasi-absolute scaling. This method can handle the extreme values, and, therefore, clinically meaningful ranges can be identified based on the distribution of the clustering variables (ranges of the quasi-absolute scaling were as follows: for digit span backward 0-2-severe (problems/impairment), 3-moderate, 4-mild, and 5-8few or none; for WCST T < 34 moderately severe, T = 35-44mild, 45 < T few or none; for the Stroop task T = 32-44 mild to moderate, T = 45-54 few or none, 55 < T above average) [36]. After conducting the hierarchical cluster analysis, we performed a K-means cluster analysis, which improves the obtained cluster solution by the relocation of cases. It starts from the initial classification and moves cases from one cluster to another if this leads to a reduction in the total error sum of squares of the cluster solution. In this method the "bad-fitting" cases are moved to other "better-fitting" clusters; thus, more homogeneous groups can be obtained. By reason of having clusters of different sample sizes, the differences across the final clusters in age and in hyperactiveimpulsive and inattentive symptoms were analyzed with the robust Welch test of equality of means. Most of the analyses were performed in SPSS 17.0, but ROPStat ([37] for details see http://www.ropstat.com) was also applied for obtaining special pattern-oriented algorithms and features.

3. Results

3.1. The Executive Functions Clusters. The attributes for clustering participants were the three factors of executive functions: inhibition (interference score on the Golden Stroop), shifting (perseverative errors on the WCST), and WM/updating (digit span backward). We obtained six clusters explaining 78.03% of the variance (considering error sum of squares). The Silhouette coefficient (the Silhouette coefficient is an indicator of good cluster cohesion and separation, and it ranges between -1 and 1. Values greater than .5 indicate reasonable partitioning of data) of the cluster structure was .793. (For the detailed demographic and behavioral properties of the clusters see Table 1.)

The first cluster consists of well-performing participants (n = 26, 30.6%) of the whole sample), so we called that none or few symptoms group. In the second cluster (n = 8, 9.4%) the performance of the children is moderately low on inhibition and mildly low on shifting. The third cluster (n = 11, 12.9%) is characterized by moderate to severe shifting impairment and moderately low working memory achievement. The fourth (n = 15, 17.6%) and sixth (n = 13, 15.3%) clusters consist of participants with severe WM problems which is associated with severe shifting impairments in cluster 6. On the other hand cluster 5 (n = 12, 14.2%) contains 12 children with moderately low WM capacity (for the detailed profiles see Figure 1).

Considering the properties of the six clusters (see Table 1) we can identify two groups with mostly TD children (none or few symptoms, moderate WM), two groups with mostly or

TABLE 1: Demographic and behavioral properties of the six clusters.

		Clusters							
		None or few	Mod Inh-Mild Shift	Mod/Sev Shift-Mod WM	Sev WM-Mild Shift	Mod WM	Sev WM-Sev Shift		
Diagnosis	TD	20 (76.9%)	5 (62.5%)	7 (63.6%)	1 (6.7%)	10 (83.4%)	0 (0%)		
	ADHD	6 (23%)	3 (37.5%)	4 (36.4%)	14 (93.3%)	2 (16.7%)	13 (100%)		
Gender	Girls	4 (15.4%)	3 (38%)	2 (18.2%)	0 (0%)	1 (9%)	1 (7.7%)		
	Boys	22 (84.6%)	5 (62%)	9 (81.8%)	15 (100%)	11 (91%)	12 (92.3%)		
Age	Mean (SD)	11.5 (1.1)	11.2 (1.3)	12.6 (.9)	10.8 (2.1)	11.2 (.9)	10.8 (1.5)		
Inattentive	Mean (SD)	6.04 (5.4)	9.63 (5.34)	10.91 (8.92)	13.93 (7.85)	5.55 (5.11)	15.55 (2.38)		
Hyperactive-impulsive	Mean (SD)	4.46 (5.94)	8.75 (8.61)	7.09 (6.53)	13.86 (8.43)	2.91 (4.41)	13.36 (4.46)		



FIGURE 1: Neuropsychological profiles of the six clusters. Homogeneity coefficient (HC) is the average of the pairwise distances within a cluster. Larger values indicate more heterogeneous clusters. Inh: inhibition, Mod: moderate, Sev: severe, Shift: shifting.

solely children with ADHD (severe WM with mild shifting, severe WM with severe shifting), and two clusters with mixed samples (moderate inhibition with mild shifting, moderate to severe shifting with moderate WM). These last two groups could be named as subthreshold or subclinical clusters. In every group there are more boys than girls (this is an attribute of the whole sample), and this ratio is not different between the clusters, $\chi^2(5) = 7.46$, P > .05. The clusters differ in age, W(5, 29.98) = 4.18, MSE = .92, P < .05, but regarding the Games-Howell post hoc test, only the third group members (moderate to severe shifting and moderately low WM) are older than the others (P < .05), except for the second cluster (moderately low inhibition and mildly low shifting).

3.2. Cognitive and Behavioral Profiles. The cluster analysis revealed different cognitive neuropsychological profiles of the

sample. Then we investigated whether these profiles match the behavioral dimensions, and we analyzed the effect of frequent comorbid syndromes as conduct disorder (CD) and learning disorder (LD).

The cognitive clusters differ both in ADHD-RS inattention, W(5, 28.26) = 13.82, MSE = 41.78, P < .01, and in hyperactive-impulsive, W(5, 28.27) = 8.39, MSE = 50.2, P < .01, scales. Considering the Games-Howell post hoc tests, the two TD-like groups (none or few symptoms, moderate WM) have lower rating scale score than the two ADHD-like groups (severe WM with severe shifting, severe WM; in each case P < .05; see Table 1) but do not differ from one another or from the two subclinical clusters. The same pattern can be observed in the hyperactive-impulsive scale: the two TDlike clusters differ significantly (P < .05) only from the two ADHD-like groups.

			Comorbie	d diagnosis		
Clusters	CD			LD		
	п	%	Adj. residual	п	%	Adj. residual
None or few	1	3.85	-2.72	2	7.69	-2.29
Mod Inh-Mild Shift	2	25	.19	0	0	-1.65
Mod/Sev Shift-Mod WM	3	27.27	.42	0	0	-1.97
Sev WM-Mild Shift	6	40	1.81	6	40	1.66
Mod WM	2	16.67	51	1	8.33	-1.34
Sev WM-Sev Shift	5	38.46	1.56	11	84.62	5.64

TABLE 2: Comorbid diagnoses in the cognitive clusters.

Note. LD = learning disorder, CD = conduct disorder. Adjusted residuals: the residual for a cell (observed minus expected value) divided by an estimate of its standard error. The resulting standardized residual is expressed in standard deviation units above or below the mean. When the absolute value of the residual is greater than 2, it can be concluded that the given cell had contribution to the chi-square result.

The clusters are different in the ratio of comorbid CD, $\chi^2(11.18)$, P < .05, and LD, $\chi^2(34.23)$, P < .01 indicated by Fisher's exact test (see Table 2). The proportions of additional CD and LD are lower in the none or few symptoms cluster than in the others. The ratio of comorbid LD is lower in the cluster with moderate to severe shifting achievement with moderate WM, but it is higher in the cluster with severe WM and shifting problems compared to the other four.

4. Discussion

In this study, we used a person-oriented statistical approach to understand the heterogeneity of EF in school-age children with or without ADHD. As we predicted, the threefactor model [11] was useful to segment children both in referred and nonreferred samples. Six different clusters were identified, where two were TD-like, two were ADHD-like groups, and two represented subthreshold categories. The executive factors were not totally independent in our sample: although shifting and updating composed separate clusters, the cognitive impairments were not severe. Interestingly, the updating factor was the most relevant in our obtained structure. While shifting could modulate the separation, inhibition had only a limited contribution to our model.

A previous research [21] presented a simpler cluster structure with three groups: children with poor inhibitory control, poor set-shifting/speed, and intact task performance. Our model was more elaborated with six clusters, which could reflect two differences between the studies. First, we measured WM, which is one of the most often found EF impairments in the developmental psychiatric literature [6]. Second, the previous study reported data from clinical sample only [21]. Another cluster analytic study [7] presented four clusters in a preschooler sample, where two represented clinical or at risk groups, while the two others showed neither ADHD risk nor EF deficits. Our cluster structure had two other groups with subclinical characteristics, in line with other studies which demonstrated mild EF deficits in nonclinical samples [13, 16].

Our clusters also varied in the severity of cognitive impairment. This result was also reflected by the latent class analysis (LCA) studies which reported different symptom severity classes in ADHD and in TD samples as well [38-45]. Our results are in line with the study of Hudziak et al. [41], where severe behavioral symptoms existed only in the clinical setting; however, mild and moderate ones were in the TD sample as well. In accordance with the LCA studies, our cluster analytic approach supports more the continuum models rather than the categorical ones in regard to symptom severity. No rigorous borders are hypothesized in a continuum model, and only the number of symptom dimensions (hyperactivity, inattentive behavior, and impulsivity) affects the ADHD taxonomy to a large extent. LCA studies often suggest subtypes like impulsive behavior without hyperactivity, daydreaming, or excessive-talkative communication style. Importantly, these studies could also identify subthreshold groups with milder symptoms in epidemiological samples. LCA is part of the person-oriented statistical methods, whose approach is paraphrased by Bergman et al. [46]. Personoriented methods (like hierarchical clustering, configural frequency analysis, latent structure models, and dense point analysis) can handle the phenomenon of symptom instability. The dynamic changes between ADHD and EF and their infinite variety may manifest in different profiles or clusters as types [47]. By using various forms of cluster analysis there is a beneficial way to test the similarity in a person-oriented way [46]. Nevertheless, in a therapeutic approach the personal focus could be relevant; however, group comparisons cannot serve this purpose.

We have hypothesized that the atypical neuropsychological clusters are differently characterized by ADHD symptoms. However, there was no difference between our clusters in the type of the symptoms ratings; that is, inattention and hyperactivity-impulsivity scales did not differ. The cognitive clusters and the behavioral dimensions only partially match in our sample. Nevertheless, both the cognitive neuropsychological and the behavioral rating scales were sensitive to symptom severity. Our clusters describe a cognitive dimension of the ADHD-TD continuum, where different types emerge from multiple EF components. This is in accordance with the transition from models of a single core deficit to multiple-deficit models that represents a paradigm shift in the way that the neuropsychology of ADHD is conceptualized. According to these changes, theoretical models emerged that attempted to account for the neuropsychological heterogeneity of ADHD [9, 18].

Comorbidity alone cannot explain the differences between the clusters. At the same time, we could see that comorbidity can transform the clusters' structure as it was previously demonstrated [7, 21, 43]. Considering that associations between ADHD-related comorbid symptoms and EF factors are not completely known [7], we should mention that in our study CD did not occur more frequently in any of the atypical cognitive clusters, and LD was obviously present only in the most severely impaired group (severe WM with severe updating). The difference in comorbid LD between our ADHD clusters could be a measurement bias due to a theoretical and cognitive overlap between the two diagnostic terms (i.e., LD and ADHD) [48]. It is still unclear whether the often reported learning problems in ADHD are parts of the core deficits or are caused by independent although cooccurring impairments in learning. Moreover, it is also possible that some children's symptoms of ADHD are secondary, caused by primary learning problems [48]. Therefore, conducting a similar cluster analytic study without comorbid LD would be important to understand this problem described above.

The three tasks used in this study are widely known and accessible for practitioners [9], which may ease implementing these findings in clinical practice (e.g., diagnostics and treatment). The Stroop task was repeatedly used to indicate the inhibition factor [10, 14]. The Wisconsin Card Sorting Test was originally proposed as a Shifting Paradigm [11]. Meanwhile, the digit span is analogous to many previously used verbal WM tasks in this line of literature [11, 13]. While our task selection was based on the three-factor model of EF [11], we would like to note that our results cannot be considered as an attempt to validate this model in ADHD. Due to the robustness of the model, we may assume that the present cluster structure would generally hold up; however, we do not expect exactly the same structure with different type of inhibition (Go/No-go, stop-signal), shifting (trail making, verbal fluency), or updating (operation span, spatial span) tasks. As another limitation, we should mention the relatively high male ratio in our sample. While the prevalence of ADHD is higher in boys than in girls, many authors suggest that epidemiological designs could balance this skewed pattern [49]. The relatively small number of participants in our clusters necessarily raises the question of reliability and generalizability of our findings. Despite the good clustering scores (ESS, HC, Silhouette) conducting a similar analysis on a larger sample is an important future task.

5. Conclusions

The behavioral ratings and the cognitive neuropsychological profiles of ADHD converged only in the dimension of symptom severity. In regard to the qualitative meaning of the clusters, the two types of information were mismatching, and there were no systematic relations among inattention, hyperactive-impulsive behavior, and the three components of executive functions. It is possible that atypicalities in EF may be manifested in nonspecific everyday problems (e.g., difficulties with turn taking in conversations, forgetfulness, losing things, etc.), similar to the comorbid learning disorder (e.g., listening or paying attention, doing math, etc.), which appeared more likely in more severe neuropsychological forms of ADHD. However, neuropsychological profiles are unique and not replaceable with rating scales. We suggest that diagnostic description of behavior needs to be quantifiable and testable, taking into consideration both the cognitive neuropsychological and the behavioral profiles of symptomatology. We also propose adopting person-oriented statistical methods in neuropsychological studies, because those are more informative in developmental research than in the variable-oriented statistics.

Conflict of Interests

The authors declare that there is no conflict of interests regarding the publication of this paper.

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