

Studi sperimentali

Metacognitive and emotional/motivational executive functions in individuals with autism spectrum disorder and attention deficit hyperactivity disorder: preliminary results

Funzioni esecutive metacognitive ed emozionali/motivazionali nel disturbo dello spettro dell'autismo e nel disturbo da deficit di attenzione con iperattività: risultati preliminari

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SUMMARY. Background. Deficits in executive functions (EF) are frequently observed in autism spectrum disorder (ASD) and in attention deficit hyperactivity disorder (ADHD). The aim of this study was to evaluate executive performances of children with ASD and ADHD, and then make between-group comparisons as well as comparisons with a control group. **Methods.** A total of 58 subjects were recruited, 17 with ASD but without intellectual impairment, 18 with ADHD-combined presentation and 23 with typical development, matched on gender, chronological age and intellectual level. They were tested on some EF domains, namely planning, mental flexibility, response inhibition and generativity, which account for both metacognitive and emotional/motivational executive functions. **Results.** Results showed a large overlapping of EF dysfunctions in ASD and ADHD and were not indicative of the presence of two real distinct EF profiles. Nevertheless, in ADHD, a more severe deficit in prepotent response inhibition (emotional/motivational EF) was found. **Conclusions.** Results are partially consistent with those found in the literature. Further studies with larger samples are needed to determine how ASD and ADHD differ in terms of their strengths and weaknesses across EF domains.

KEY WORDS: mental flexibility, planning, inhibition, generativity.

RIASSUNTO. Scopo. I deficit delle funzioni esecutive (FE) sono frequentemente osservati nei disturbi dello spettro dell'autismo (ASD) e nel disturbo da deficit di attenzione con iperattività (ADHD). Lo scopo di questo studio è quello di valutare e confrontare le funzioni esecutive metacognitive ed emozionali/motivazionali di bambini con ASD e ADHD, sia fra di loro che con un gruppo di controllo. **Metodi.** Il campione è costituito da un totale di 58 soggetti, di cui 17 con ASD senza disabilità intellettiva, 18 con ADHD-manifestazione combinata e 23 con sviluppo tipico, abbinati per genere, età cronologica e livello intellettivo. Le valutazioni hanno riguardato alcune aree del funzionamento esecutivo, nello specifico pianificazione, flessibilità mentale, generatività e inibizione della risposta, che rappresentano sia le funzioni esecutive metacognitive sia quelle emozionali/motivazionali. **Risultati.** I risultati hanno rilevato un'ampia sovrapposizione delle disfunzioni esecutive nei due gruppi clinici con ASD e ADHD, e non sono stati indicativi della presenza di due profili realmente distinti del funzionamento esecutivo. Tuttavia, nell'ADHD è stato trovato un deficit più severo nell'inibizione della risposta prepotente (funzione emozionale/motivazionale). **Conclusioni.** I risultati del nostro studio sono parzialmente in accordo con quelli della letteratura. Ulteriori ricerche con gruppi più numerosi potranno chiarire più approfonditamente quali punti di forza e debolezza, nell'ampio spettro delle funzioni esecutive, differenzino fra loro le prestazioni di persone con ASD e ADHD.

PAROLE CHIAVE: flessibilità, pianificazione, inibizione, generatività.

INTRODUCTION

The term “executive functions” (EF) refers to mental processes, mainly regulated by frontal lobes, that are necessary to maintain effective goal-directed behavior¹ and dy-

namic behavior self-regulation². EF include different abilities, such as planning, working memory, mental flexibility, response initiation, response inhibition, impulse control and action monitoring³. Two major types of EF have been suggested by Ardila⁴: a) metacognitive EF, typically measured

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by using neuropsychological tests and relating to activities of the dorsolateral prefrontal cortex (this latter having a role in planning, abstracting, problem solving, and working memory); b) emotional/motivational EF, which are accountable for coordinating cognition and emotion throughout socially accepted strategies (“inhibitory control” of behaviors), with neural correlates residing in the orbitomedial prefrontal cortex. Two major types of executive dysfunction syndromes, namely metacognitive and motivational/emotional, were identified by Ardila⁴, the first one referring to impairments in intellectual abilities, and the second relating to impairments in behavioral control.

Impaired EF are frequently observed in neurodevelopmental disorders, including autism spectrum disorder (ASD) and attention deficit hyperactivity disorder (ADHD). Based on DSM-5 criteria⁵, ASD is a severe neurodevelopmental disorder characterized by impairment in social communication and interaction across multiple contexts, and restricted and repetitive patterns of behaviors, interests, or activities, which cause clinically significant impairment in social, occupational, or other important areas of current functioning; ASD could present with or without intellectual impairment (low functioning ASD or high functioning ASD, respectively). In addition to the core features, deficits in EF have been widely reported in ASD, especially characterized by deficient planning, flexibility and behavior regulation in ecological contexts⁶. ADHD is a neurodevelopmental disorder characterized by a persistent pattern of inattention and/or hyperactivity-impulsivity that interferes with functioning or development; symptoms are present in two or more settings; individuals with ADHD can be further divided into those who show problems in all the aforementioned domains (combined presentation), those primarily evidencing attention problems (predominantly inattentive presentation) and those with mostly hyperactive and impulsive symptoms (predominantly hyperactive/impulsive presentation). Substantial EF deficits in individuals with ADHD have been reported, especially in response inhibition and vigilance as well as in working memory, planning and flexibility⁵.

Only few studies have compared EF across ADHD and ASD groups (Table 1)⁷⁻¹⁸: although response/motor inhibition, sustained attention and working memory were found to be more severely impaired in ADHD, while flexibility and planning more severely impaired in ASD, the hypothesis of a double dissociation has not been consistently demonstrated across studies; controversial results might be due either to task selection, participant matching (especially on IQ and gender) or ability level. Further studies comparing ASD and ADHD children are needed to determine how these two disorders differ in terms of their strengths and weaknesses across EF domains.

This study aimed to compare EF profiles of children with ASD and ADHD, matched on gender, chronological age, and intellectual level. The following EF domains have been investigated: planning, mental flexibility, response inhibition and generativity, as described by Hil¹⁹, who derived them from the analysis of studies with well-matched control groups; the same domains were later investigated by Robinson et al.³. These domains refer to metacognitive EF (planning, mental flexibility and generativity) and emotional/motivational EF (response inhibition). Based on literature data, we hypothesized that ADHD and ASD wouldn't show two

distinct EF profiles, and that quantitative differences in some EF domains would be found, with response inhibition being more globally and severely impaired in ADHD children, while flexibility and planning being more deficient in ASD children. As far as generativity is concerned, very few studies are present in the literature, often with inconsistent results; therefore, our analysis on generativity was exploratory in nature.

METHODS

Participants

Fifty-eight children participated in the study. Subjects were recruited in a centre highly specialized in diagnosis and treatment of developmental disorders. Participants with ASD were diagnosed by a multidisciplinary team of professionals, based on DSM-5 criteria⁵. Diagnoses were further confirmed using the autism diagnostic interview-revised (ADI-R)²⁰ and the autism diagnostic observation schedule (ADOS)²¹. Participants with ADHD were also diagnosed by a multidisciplinary team of professionals, based on DSM-5 criteria⁵. A T-score >60 for at least one of the ADHD-related indexes of Conners' Parent Rating Scale-Revised-Long Version (CPRS-R:L)^{22,23} was used to further confirm diagnoses.

The first clinical group was made of 17 individuals with ASD (15 males and 2 females), IQ ≥ 80 (IQ range: 80-134) and mean chronological age 9.5 years (SD=3.27). The second group was made of 18 individuals with ADHD-combined presentation (15 males and 3 females), IQ ≥ 80 (IQ range: 80-121) and mean chronological age 9.6 years (SD=2.43). The control group was made of 23 individuals (19 males and 4 females) with typical development (TD) and mean chronological age 9.9 years (SD=2.65), recruited from local public schools. All participants from the three groups were matched on gender (Chi Square test, $p=0.91$), chronological age (Kruskal-Wallis ANOVA test: $H=0.799$, $p=0.67$) and performances at the Italian version of Raven's Colored Progressive Matrices-CPM²⁴ (Kruskal-Wallis ANOVA test: $H=5.47$; $p=0.08$), a non-verbal test of general intelligence. We chose to use CPM due to their being independent from language, reading or writing skills, and to the simplicity of their use and interpretation; furthermore, CPM are included, as a measure of intelligence, in the national Guidelines for the Diagnosis and Pharmacological Treatment of ADHD (http://www.iss.it/binary/wpop/cont/SINPIA_L.g.ADHHD.1116940207.pdf) and in the regional Guidelines for educational services implementation for persons with ASD (www.gurs.regione.sicilia.it/Gazzette/g07-09/g07-09-p14.html). Parental informed consent was obtained for each participant included in the study. Ethical approval was granted by the local Ethical Committee.

Measures

The following testing instruments were used to assess the different domains of EF:

- Planning: *Tower of London (ToL)*, version included in the BVN 5-11²⁵ or BVN 12-18²⁶, two Italian neuropsychological assessment batteries for children aged 5-to-11 or 12-to-18 years; Clock Drawing Test (CDT), version included in the Italian Brief Neuropsychological Evaluation (ENB)²⁷.
- Mental flexibility: Wisconsin Card Sorting Test (WCST), Italian version²⁸.

- Response inhibition: Stroop Test, Italian version²⁹; Go/No-Go trial, from the Frontal Assessment Battery (FAB)³⁰.
- Generativity: Verbal fluency tasks (category and phonemic) from the BVN 5-11²⁵ or BVN 12-18²⁶.

Procedures

Tests were administered by a clinical psychologist and presented in a pre-established order in one single session. The pre-established order presentation of tests was not expected to affect children's responses. Although there might have been an influence of the order presentation, it has been the same for all the participants, not affecting the validity of comparisons between groups.

Statistics

The Kolmogorov-Smirnov and Lilliefors tests were used to test whether data relating to all variables considered in the study derived from a normally distributed population. Since not all the variables were normally distributed, non parametric statistical tests were used for the comparisons between groups, and namely: comparisons between multiple groups (ASD-ADHD-TD) were carried out using the Kruskal-Wallis ANOVA test; comparisons between two groups (ASD vs. ADHD; ADHD vs. TD; ASD vs. TD) were carried out using the Mann-Whitney's U test.

RESULTS

Results obtained, shown in Table 2, are described in separate sections, one for each EF function investigated.

Planning

Statistically significant differences across the three groups were found both at ToL and Clock test. No difference emerged from the comparison between ASD and ADHD; when compared to TD, ASD showed a statistically significant differences at both measures, whereas ADHD at Clock test only.

Generativity

The comparison between the three groups showed statistically significant differences both in category and phonemic fluency. No difference was found in the comparison between ASD and ADHD. In the comparison with TD group, ADHD individuals showed a statistically significant difference both in Category and Phonemic Fluency, whereas ASD subjects only in the Category Fluency.

Inhibition

At the Go/No-Go test, a statistically significant difference was found in the comparison between the three groups. No difference was found between ASD and ADHD, whereas both ASD and ADHD showed a statistically significant difference in the comparison with TD. At the Stroop word-color test, no statistically significant difference was found be-

tween the three groups as for speed, unlike for accuracy (number of errors), in which a statistically significant difference was also found in the comparison between ASD and ADHD; when compared to TD, a statistically significant difference was found only for ADHD.

Flexibility

Statistically significant differences were found in all the WCST parameters when comparing the three groups: no difference was found between ASD and ADHD, whereas statistically significant differences emerged when ADHD was compared to TD on all the WCST parameters. ASD-TD comparisons showed statistically significant differences only in some of the parameters above, and namely in number of errors, perseverative responses and errors, and failure to maintain set.

CONCLUSIONS

This study aimed to investigate some EF domains, such as planning, mental flexibility, generativity and response inhibition in children with ASD and ADHD-combined presentation, with the aim of confirming the presence of differences in types and severities of EF deficits. In our sample, largely overlapping EF deficits were found in ASD and ADHD individuals, with impairments both in motivational/emotional and metacognitive EF⁴; all investigated EF domains appeared to be impaired and, consistently with our general hypothesis, results did not suggest the presence of two distinct EF profiles in individuals with ADHD and ASD. Only the parameter "number of errors" at Stroop test differentiated ADHD from ASD, thus suggesting control of prepotent response (a sub-category of response inhibition) being impaired in ADHD and relatively preserved in ASD. This is likely to be a typical characteristic of executive dysfunctioning (motivational/emotional type) in ADHD, as shown in some literature studies^{9,11,13,18}; on the contrary Johnson et al.¹² did not detect statistically significant differences in response inhibition between the two clinical groups.

With regard to planning, a global impairment was found in ASD when compared to TD (both ToL and Clock tests), but no statistically significant difference was found in the comparison between ASD and ADHD. Contrary to the majority of literature studies^{7,9,15,17} but consistently with results by Happé et al.¹¹, in our study, planning deficits did not appear to be a feature that clearly differentiates ASD from ADHD individuals. With regard to ADHD, some studies showed preserved planning abilities^{8,10,16,18}, whereas planning turned out not to be entirely preserved in our sample: indeed, ADHD scored lower in Clock test than TD, with a statistically significant difference. This result was consistent with a previous study that used the Clock test³¹, in which ADHD individuals showed more errors than TD, because of (in the opinion of the authors) their poor planning abilities; based on our results on motor response inhibition (comparison between ADHD and TD in Go/no-go test), the hypothesis that the clock drawing could have been affected also by poor motor response control seemed justified.

As far as flexibility is concerned, our initial hypothesis was not confirmed in this study: inconsistently with some lit-

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Table 1. Review of studies that have reported EF impairments in ASD and ADHD.

Authors	Sample	Working Memory	Response/Motor Inhibition	Planning	Mental Flexibility	Sustained Attention	Verbal Fluency	Conclusive Hypothesis
Pennington and Ozonoff ¹	Children	ASD NI in ADHD	ADHD NI in ASD					Dissociation
Sergeant et al. ⁷	Children		ADHD NI in ASD	ASD<ADHD	ASD<ADHD		ADHD	
Gargaro et al. ⁸	Children		ADHD NI in ASD	ASD NI in ADHD	ASD NI in ADHD	ADHD NI in ASD		Double dissociation
Geurts et al. ⁹	Children n. 136	NI in ASD	ADHD<ASD	ASD<ADHD	ASD<ADHD		ADHD<ASD	
Goldberg et al. ¹⁰	Children n. 70	ADHD<ASD	NI in both ASD and ADHD	NI in both ASD and ADHD	NI both in ASD and ADHD			
Happé et al. ¹¹	Children and adolescents n. 94	ADHD<ASD	ADHD < ASD	No difference between ASD and ADHD	No difference		No difference	Less severe and persistent EF deficits in ASD
Johnson et al. ¹²	Children n. 62		No difference			ADHD<ASD		
Sinzig et al. ¹³	Children and adolescents n. 70	ADHD<ASD	ADHD<ASD	ASD<ADHD	ASD<ADHD			
Corbett et al. ¹⁴	Children n. 54	ASD<ADHD	ASD<ADHD		ASD<ADHD			More pronounced EF impairments in ASD
Semrud-Clikerman et al. ¹⁵	Children and adolescents n. 96		Only ecological measures impaired, but no difference between ASD and ADHD	ASD<ADHD	Only ecological measures impaired, but no difference between ASD and ADHD			
Miranda-Casas et al. ¹⁶	Children		ADHD NI in ASD	ASD NI in ADHD	ASD NI in ADHD	ADHD<ASD		Double dissociation
Salcedo-Marin et al. ¹⁷	Children and adolescents n. 103			ASD<ADHD (ecological measure)				
Paloscia et al. ¹⁸	Children N. 79	ASD<ADHD	ADHD<ASD NI in ASD	NI in both ASD and ADHD	ASD<ADHD			More pronounced EF impairments in ASD

ASD= Autism Spectrum Disorder; ADHD= Attention Deficit Hyperactivity Disorder; <= lower ability; NI= non impaired.

erature data^{7,9,13,14,18}, which show flexibility as being more markedly impaired in ASD than in ADHD individuals, in our study perseverative responses and errors turned out to be common features in both ASD and ADHD; according to results obtained by Happé et al.¹¹, no statistically significant differences were found in the comparison between ASD and ADHD children, and flexibility was found to be more globally impaired in ADHD (all the WCST parameters were impaired in the comparison with TD).

Results on flexibility were confirmed by scores on generativity; indeed, verbal fluency tasks measure flexibility in generating items and also require response inhibition to control errors¹¹; in line with the literature^{7,9}, in our study ADHD children showed overall poorer performances, revealing impairments in both category and phonemic fluency when compared to TD, whereas ASD children showed impairments only in the fluency category.

This study has some aspects that may be considered as

Table 2. Comparison between ages and the different test results obtained from ASD, ADHD and TD children in this study.

	1. ASD (n=17)	2. ADHD (n=18)	3. TD (n=23)	Kruskal-Wallis ANOVA	Mann-Whitney U test		
	median (25-75%)	median (25-75%)	median (25-75%)		1 vs. 2	1 vs. 3	2 vs. 3
				p<	p<	p<	p<
Age, years	9.2 (7.1-10.9)	9.25 (7.9-10.4)	10.3 (7.5-12.1)	NS			
Colored Progressive Matrices	26 (22-30)	26 (22-29)	30 (24-33)	NS			
Tower of London	6 (5-6)	6 (6-7)	7 (6-9)	0.0088	NS	0.005	NS
Clock Drawing Test	6.5 (0-8.5)	4 (1-9)	9.5 (7.5-10)	0.0013	NS	0.0017	0.0037
Category Fluency	35 (25-45)	33 (30-40)	46 (41-59)	0.0072	NS	0.014	0.0046
Phonemic Fluency	20 (14-24)	14 (10-18)	23 (20-30)	0.0029	NS	NS	0.0006
Go/no-go test	3 (2-3)	2 (2-3)	3 (3-3)	0.0003	NS	0.033	0.00045
Stroop test: time, s	74 (55-89)	91 (57.5-110.5)	58 (53-70)	NS			
Stroop test: errors, n	2 (0-5)	6 (3.5-6.5)	1 (0-2)	0.0002	0.012	NS	0.00005
WCST: trials, n	107 (87-128)	128 (114-128)	91.5 (71-122)	0.028	NS	NS	0.0096
WCST: errors, %	28 (21-37)	29 (25.5-37.5)	19 (12-27)	0.002	NS	0.0074	0.0013
WCST: perseverative responses, %	24 (13-26)	18 (13-21)	9 (7-13)	0.0001	NS	0.0002	0.0009
WCST: perseverative errors, %	18 (10-32)	20 (17-24)	7.5 (5-17)	0.0039	NS	0.041	0.00055
WCST: completed categories, n	6 (3-6)	5 (3-6)	6 (6-6)	0.034	NS	NS	0.021
WCST: failure to maintain set, n	1 (1-2)	1 (0-3)	0 (0-1)	0.01	NS	0.0096	0.026

ASD: Autism Spectrum Disorder; ADHD: Attention Deficit Hyperactivity Disorder; TD: Typical Development; WCST: Wisconsin Card Sorting Test.

limitations. Children from all the three groups were matched on their intellectual levels; this very important criterion has been frequently neglected in previous studies of the literature¹¹; we decided to use only one measure (the Raven's CPM), nevertheless, the use of more comprehensive measures, such as the Wechsler scales, are more likely to guarantee a better IQ-based matching.

Another limitation is that the ADHD group showed only a combined presentation: therefore, caution must be taken in generalizing results to the other group presentations (e.g., predominantly hyperactive/impulsive or predominantly inattentive). Also, the relatively small sample size probably explains why some findings are inconsistent with the literature data, although they certainly need to be confirmed by studies with larger sample sizes.

Conflict of interests: the authors declare they have no competing interests.

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