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Cognitive flexibility and planning processes in Autism spectrum disorder and Attention deficit/hyperactivity disorder

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ABSTRACT

Background: ADHD is a neurodevelopmental disorder which is often in comorbid with other disorder, such as autism spectrum (ASD), hyperactive-impulsive subtype, visual attention deficits and impulsivity. The aim of this research is to underline the differences in the three psychopathological profiles and to investigate whether and how comorbidity with hyperactive syndrome can influence executive performances in children with autistic spectrum disorder.
Methods: Participants in the current study were 60 children aged between 5 and 10 years, equally divided into 4 groups: those with typical development, with high-functioning autism, autism with comorbid ADHD and ADHD. The psychological instruments regard Raven's Colored Progressive Matrices (RPCM), Tower of London (TOL), Modified Card Sorting Test (MCST).
Results: ASD+ADHD group shows worse performance in terms of planning than the other groups involved in the research. The presence of hyperactivity in children with ASD makes cognitive flexibility even worse. ASD+ADHD group has worse executive functioning than children with only ADHD.
Conclusions: This study found that a comorbidity ASD+ADHD makes executive functioning worse in children with ASD, which is already impaired due to the characteristics of the syndrome itself.

Keywords: Autism spectrum disorder; Attention-deficit/hyperactivity disorder; executive dysfunctions, perseverative response.

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Introduction

Autism spectrum disorder (ASD) and attention deficit/hyperactivity disorder (ADHD) are a group of neurodevelopmental disabilities that appear in childhood and are associated with a wide range of difficulties across different domains. The ADHD was characterized by attention, hyperactivity, and impulsivity deficits. The ASD was defined by impaired social functioning, limited communication skills and stereotyped patterns of behaviour (American Psychiatric Association, 2013). The comorbidity of ADHD in ASD (ASD + ADHD) ranges from 14% to 70% (Antshel & Russo, 2019) and pose serious challenges to clinical practice (Chen et al., 2021). Despite the unique symptom features, both ASD and ADHD had some overlapped cognitive impairments (Wang et al., 2018). In the current literature there is a debate about the similarities and differences of cognitive functions between ADHD and ASD. More study underlines the role of the theory executive dysfunction to understand cognitive impairments in ADHD (Caprì, Santoddì & Fabio, 2020; Holmes et al., 2010) and ASD (Panerai et al., 2014; Fabio et al., 2020).

Executive functions (EF) are a complex cognitive control process, which permit the self-regulation and self-direction of a behaviour towards a goal, the modification of a behaviour in the light of new information, the ability to solve novel problems and the decision making and evaluation of risks through the planning of future, the identification of prioritises and sequences actions (Miyake & Friedman, 2012; Bierman & Torres, 2016). According to Zinke and colleagues (2010) EF includes different functions. Planning refers to the ability to formulate actions in advance and to approach a task in an organized, strategic, and efficient manner (Panerai et al., 2014; Boshomane, Pillay, & Meyer, 2020). Mental flexibility is the ability to spontaneously shift to a different action or thought in response to situational changes (Panerai et al., 2014). Response inhibition refers to the ability to suppress irrelevant stimuli or impulse to achieve a goal (Mokobane et al, 2020). Moreover, previous studies have highlighted the presence of impaired automatic and controlled processes in ADHD children (Caprì et al., 2020). The latter in fact explain the lack of ability to automate and control behaviors which produce an increase in attentional resource requirements, poor resistance to interference, high sensitivity to distraction and excessive effort in carrying out tasks. Therefore, an impairment of automatic and controlled processes means, for a child with ADHD, slowness in terms of processing speed (Caprì et al., 2020; Fabio & Caprì, 2019). The centrality of the deficit in EF in ADHD children is not excluded, despite these studies. Indeed, EF impairments have been evaluated as central deficits in ADHD and ASD (Craig et al., 2016). ADHDs' symptoms arise from a deficit in specific EF domains (response inhibition, working memory, set-shifting and planning), which lead to important deficit judgement, organisation, planning, behavioural disinhibition and cognitive flexibility (Ciuluvica et al., 2013; Faraone, 2015; Boshomane, Pillay, Meyer, 2020). Moreover, in accordance with a recent study of Mansour and colleagues (2021) and Mokobane and colleagues (2020) children with ADHD report worse inhibiting response, due to symptoms of inattention.

Typical EF deficits are also present in ASD children. In fact, they show significantly impaired on planning task and on mental flexibility. These deficits cause greater perseverative responses and errors because they enact ritualistic and stereotyped behaviours. Consequently, the lack of mental flexibility, evident from increased perseverations, seemed to be a distinctive characteristic of autism. This is probably due to the fact that the mental flexibility requires multiple skills (production of rules, working memory, use of feedbacks, and shifting ability) that are poor in ASD children (Mari et al., 2003; Hill, 2004). Moreover, children with autism shown usually unimpaired response in inhibition of a prepotent response and control of interfering stimuli (Christ, Holt, White, & Green, 2007; Panerai et al., 2014). For this reason, these deficits might be assumed as typical feature of ASD executive profile; this were characterized by deficient planning and impaired mental flexibility, which was associated with decreased adaptive abilities, behaviour regulation impairment, communication difficulties and facial emotional recognition deficits (Fabio et al., 2020).

Previous studies underlined that executive dysfunctions have different expression in ADHD and ASD (Demetriou, DeMayo, & Guastella, 2019; Demetriou, Lampit, Quintana et al., 2018). In accordance with Craig and colleagues (2016) and Unterrainer and colleagues (2020), ASD children mostly manifested an alteration in planning (worse performance and more planning times), whereas ADHD symptoms would affect inhibition response. In particular, children with ASD-only employ more time in initial planning times, compared to ADHD and TD children.

Conversely, ASD+ADHD children showed negligible differences in planning duration. An explanation may be that the ADHD symptomatology with ASD do not have a detrimental impact, because the two disorders do not add up (Unterrainer et al., 2016). Thus, it remains unclear whether the executive function problems attributed to ASD are truly due to shared liability or simply reflect comorbid ADHD symptoms.

Furthermore, children with ASD show difficulties in the correct placement of stimuli (categories achieved; CA), a greater number of errors in misclassifying according to a previous sorting rule, despite receiving negative feedback (perseverative errors; PE), and random errors (non-perseverative errors; NPE). All these types of errors are related to the inability to inhibit a predominant response (Landry & Mitchell, 2021; Wang et al., 2018) and reflect executive dysfunction (Sullivan et al., 1993). In contrast, children with ADHD report greater difficulty in changing response strategies despite the rule remaining the same (maintain set errors; FMS). This type of error reflects an attentional deficit (Sullivan et al., 1993). Children with ASD complete fewer categories (CA) than typical development (TD) children (Bennetto, Pennington & Rogers, 1996; Panerai et al., 2014; Kado, Sanada, Oono, Ogino & Nouno, 2020) and commit more PE than TD and ADHD children (Yasuda et al., 2014). Additional insights come from a study of Westwood and colleagues (2016). They underlined that ASD children made more PE, while children with ADHD and TD did not differ on the PE scores, but children with ADHD made more failure to maintain set errors.

On clinical practice become interesting to analyses not only how executive dysfunctions find expression in ADHD and ASD, but also the executive dysfunctions difference that occur between these disorders. The presence of ADHD-related symptoms may exacerbate cognitive problems on individual with ASD and, thus bring on a greater variety and intensity of deficit than ASD alone (Mansour et al., 2017). In accordance with Craig and colleague (2016) ASD+ADHD children show impairment with ASD children in both flexibility and planning, and they show impairment with ADHD in the response inhibition. Therefore, children with ASD+ADHD reported more poorer attention than ASD alone (Lundervold et al., 2016), probably because ADHD symptoms have an addictive effect on EF (Pascualvaca et al., 1998; Robinson, 2009; Yasuda et al., 2014), and ASD+ADHD and ASD children show more planning impairments than ADHD (Pitzianti et al., 2016). In summary, children with ASD+ADHD exhibit more impairments, followed by ASD and ADHD respectively (Benallie et al., 2021).

The literature appears to be scarce in the study on the comorbidity between ASD + ADHD. This is an important question that forms the rationale for this study. The research aim is to analyze how the executive functions manifest in children with ASD, ADHD, ASD+ADHD and TD; and underline the differences among these four groups regarding EF (planning and cognitive flexibility). And another focus is whether and how much the presence of hyperactivity affects the performance of children with ASD.

Method

Participants/Subject

The research sample comprised 4 groups of each 15 children, for a total of 60 children. Chronological ages in the sample ranged from 5 to 10 years (M = 7,50 years, SD = 1,722). The four groups are divided in: high-functioning autism (ASD-only), autism in comorbidity with Attention-Deficit/ Hyperactivity Disorder (ASD+ADHD), Attention-Deficit/ Hyperactivity Disorder (ADHD-only) and typical development (TD). Children with autism are diagnosed with the Autism Diagnostic Observation Schedule-Second Edition (ADOS-2; Colombi et al., 2013) and they are included in this study. Children with ADHD are diagnosed with ADHD Rating Scale-5 for Children and Adolescents (Marzocchi & Cavallero, 2019) were recruited from three different rehabilitation centres operating in the area of the city of Messina, which provided diagnoses for all participants following standard DSM-V (American Psychiatric Association, 2013). The control group was recruited through schools in the province of Messina. The inclusion criteria of the subgroups were: (a) a diagnosis of high-functioning autism, Autism in comorbidity with ADHD, and ADHD; (b) a full scale IQ higher than 85 in the Raven's Standard Progressive Matrices. The exclusion criteria were: (a) presence of comorbidity with others behavioral disorders, experience of any kind of sensory problems or impairments and mental retardation; (b) presence of identifiable chromosomal or neurological conditions (e.g., history of fragile X, encephalitis, or other known medical conditions associated).

Procedure and Measures

This study was performed following the recommendations of the *Ethical Code of the Italian Association of Psychology* (AIP) and all subjects were given written informed consent following the Declaration of Helsinki (2013). The protocol was approved by the Ethics Committee of the Centre for Research and Psychological Intervention (CERIP) of the University of XXXXX (protocol number: 30465). The research was conducted with the use of tests and tools for the evaluation of the variables under study, in order to arrive at a measurement as reliable as possible of the processes taken into account. Before administering the test and tools, informed consent was collected. Only children whose parents provided informed consent took part in the study. To this end, the subjects were first administered *Raven's Coloured Progressive Matrices* (CPM; Belacchi et al., 2008) with the aim to determine the IQ so that proves homogeneous in the reference sample. It was later verified the objectives of the research itself, consisting of the evaluation of the executive functions of the subjects. In this case, the test considered were the *Tower of London* (TOL; Shallice, 1982) and the *Modified Card Sorting Test* (MCST; Nelson, 1976).

The *Tower of London* (TOL; Shallice, 1982) is used to assess the ability of planning; requires the subject to perform a task only after thinking about the sequence of operations required to carry it out; assesses the skills of strategic decision and problem solving skills in children 4 to.

In particular required three operations: (a) formulate a master plan, (b) identify under goals and organize them within a sequence of movements, (c) preserve under goals and master plan in the working memory (Morris et al., 1988; Owen et al., 1990; Shallice, 1982). In fact in the TOL, coloured discs must be moved one by one from an initial state to match a goal state. Instructions are given to plan the whole sequence of moves that must be carried out mentally, before executing the sequence. TOL measures decision time (time between seeing the discs and making the first move), execution time (time spent on executing the plan), total time (trials solves in the minimum moves possible or within a specified time limit), and total point (number of moves in excess of the minimum necessary to complete the task). Poor performance on the TOL is usually interpreted as inability to plan efficiently (Shallice, 1982).

The *Modified Card Sorting Test* (MCST; Nelson, 1976) evaluates the ability of abstract reasoning and modification of cognitive strategies to changing circumstances. It is the simplified version of the *Wisconsin Card Sorting Test* (WCST; Berg, 1948; Grant & Berg, 1948) was adapted for the analysis of subjects in children aged 4 to 13 years. Version simpler and shorter administration, the MCST makes use of two sets of 24 cards instead of two to 64, being eliminated all the cards that shared more than one attribute with a card - stimulus. The subject is asked to combine the cards response to the stimulus, according to certain criteria referring to specific parameters.

The *Modified Card Sorting Test* (MCST; Nelson, 1976) measures the score of three types of error: failure to maintain set (FMS), perseverative error (PE), and non-perseverative error (NPE). An FMS error occurs when the participant changes response strategies despite the rule remaining the same. This type of error is regarded as attentional (Sullivan et al., 1993). PE refers to “the number of errors that the subjects continued to incorrectly sort according to a previous sorting rule, although they received negative feedbacks” (Wang et al., 2018). This type of error is related to a failure to inhibit a predominant response (Landry & Mitchell, 2021). NPE are generally considered to be random (Sullivan et al., 1993).

Results

Statistics and data analysis

The Statistical Package for the Social Science (SPSS 25) was used to conduct descriptive statistics and Pairwise comparison (Kruskal-Wallis test). The results are first discussed with reference to descriptive statistics, secondly with reference to Pairwise comparison.

Regarding the cognitive flexibility (table 1), the ASD-only group and ASD+ADHD, reported the same means of PE, NPE and total errors. The ADHD-only group made a slightly higher means of PE and a higher means of NPE and total errors than TD group. Conversely, the ADHD-only group reported lower PE, NPE and total errors than the ASD-only group.

MSCT	**Group**	**Mean ± SD**
PE	ASD+ADHD	41 ± ,000
	ASD-only	41 ± ,000
	ADHD-only	2,87 ± 2,696
	TD	2,20 ± 2,704

NPE	ASD+ADHD	0
	ASD-only	0
	ADHD-only	15,47 ± 8,863
	TD	7,87 ± 11,783
Total errors	ASD+ADHD	41 ± ,000
	ASD-only	41 ± ,000
	ADHD-only	18,33 ± 9,363
	TD	10,07 ± 12,115

Table 1. Descriptive Statistics (mean and standard deviation) of the scores obtained by the groups on the Modified Card Sorting Test (MCST).

Regarding the planning (table 2) the ASD-only group and ASD+ADHD, reported a different mean of decision time, execution time, total time and total point. In the ASD+ADHD group there is a very low mean compared to all the other groups considered. Conversely, about time (decision time, execution time, total time) the ADHD-only group reported a lower mean than ASD-only group and ASD+ADHD group. Also, the ADHD-only group reported better point than the ASD+ADHD group and the ASD-only group.

TOL	Group	Mean ± SD
Decision time	ASD+ADHD	,20 ± ,775
	ASD-only	250,60 ± 49,977
	ADHD-only	112,47 ± 37,709
	TD	93,93 ± 31,658
Execution time	ASD+ADHD	1,20 ± ,775
	ASD-only	251,60 ± 49,977
	ADHD-only	113,47 ± 37,709
	TD	94,93 ± 31,658
Time	ASD+ADHD	2,20 ± ,775
	ASD-only	252,60 ± 49,977
	ADHD-only	114,47 ± 37,709
	TD	95,93 ± 31,658
Point	ASD+ADHD	4,27 ± ,704
	ASD-only	6,80 ± 1,897
	ADHD-only	24,40 ± 3,888
	TD	27,33 ± 2,920

Table 2. Descriptive Statistics (mean and standard deviation) of the scores obtained by the groups on the Tower of London (TOL).

The Pairwise comparison (Kruskal-Wallis test) was reported in table 3. In particularly, the ASD+ADHD differs statistically significantly from the group with TD as regards both PE ($p < 0.00$) that of NPE ($p < 0.001$). These groups also differ in total errors ($p < 0.00$). There is also a significant difference between the ASD-only and the TD group in PE ($p <0.000$) and NPE (p <

0.001) as well as significant differences in the total errors (p < 0.00). The ASD+ADHD and ASD-only groups compared with the ADHD-only group show the same statistically significant differences in terms of PE (p < 0.00), NPE (p < 0.00) and in total errors (p <0.00). However, there are no statistically significant differences between the ASD+ADHD group and ASD-only group, regarding PE, NPE and total errors.

Groups	**MSCT**		
	PE (*p*-Value)	**NPE (*p*-Value)**	**Total errors (*p*-Value)**
ASD+ADHD / TD	.000*	.001*	.000*
ASD+ADHD / ADHD-only	.000*	.000*	.000*
ASD+ADHD / ASD-only	1.000	1.000	1.000
TD / ADHD-only	1.000	.604	1.000
TD / ASD-only	.000*	.001*	.000*
ADHD-only / ASD-only	.000*	.000*	.000*

Table 3. Pairwise comparison (Kruskal-Wallis test) of the indices obtained from the Modified Card Sorting Test (MCST). *The significance level is .05. The significance values have been corrected by the Bonferroni correction for multiple tests.

As regards the planning and in relation to the performance provided to the TOL test, in general, pairwise comparisons were made between all the groups involved, as shown in table 4. There were statistically significant differences between the ASD+ADHD group and ASD-only group (p <0.00) in execution and decision times, but there were no significant differences in the score between the two groups (p <0.460). Significant differences emerge, not only in execution and decision times, but also in the score level between the ASD+ADHD group and the ADHD-only group (p <0.00). The ADHD-only group differs from the ASD-only group in execution, decision and total time (p <0.013) and also in terms of score (p <0.007). Finally, pairwise comparisons were also carried out between the group of subjects with TD and the group of ASD-only and ASD+ADHD. There are statistically significant differences between the ASD+ADHD group and the TD group in execution, decision and total time (p <0.009) and also in the score level (p <0.00). The TD group and the ASD-only group (p <0.001) differ in the execution times, the decision times, the total time (p <0.001) and in the score (p <0.00).

Groups	**TOL**			
	Decision time (*p*-Value)	**Execution time (*p*-Value)**	**Time (*p*-Value)**	**Point (*p*-Value)**
ASD+ADHD / TD	.009*	.009*	.009*	.000*
ASD+ADHD / ADHD-only	.000*	.000*	.000*	.000*
ASD+ADHD / ASD-only	.000*	.000*	.000*	.460
TD / ADHD-only	1.000	1.000	1.000	1.000
TD / ASD-only	.001*	.001*	.001*	.000*
ADHD-only / ASD-only	.013*	.013*	.013*	.007*

Table 4. Pairwise comparison (Kruskal-Wallis test) of the indices obtained from the Tower of London test (TOL). *The significance level is .05. The significance values have been corrected by the Bonferroni correction for multiple tests.

Discussion and Conclusion

The results offer valuable insights into the cognitive profiles of the different groups in the study and shed light on the differences in mental flexibility and planning between children with TD, ASD-only, ADHD-only and ADHD+ASD.

Regarding the planning, there are significant differences between groups. There are differences concern the decision and execution times that are longer in ASD-only group, while these times are significantly reduced in TD group. The total scores obtained in the *Tower of London* test are also found to be lower in ASD-only group. These results confirm the literature. It shows that ASD-only children show impaired planning, with long planning times and worse performance (Craig et al., 2016; Unterrainer et al., 2020). It was important to carry out a comparison between ADHD-only group and ASD-only group to understand if there were significant differences in execution and decision times. The results reported that ADHD-only group performs reduces decision and execution times. This is due to the presence of the component of impulsivity and to the presence of deficit in EF domains (response inhibition, working memory, set-shifting and planning), that is characterizing the disorder (Polanczyk et al., 2007; Ciuluvica et al., 2013; Faraone, 2015; Boshomane, Pillay, Meyer, 2020). Moreover, this study evaluated if the comorbidity of ADHD in children with ASD could lead to a deterioration in executive functions. The comorbidity seems to significantly worsens the performance. In particular, ASD+ADHD group have much shorter decision and execution times and a worse score than children with ADHD-only. In fact, the test explicitly highlights the "impulsive" factor typical of comorbidity with ADHD that pushes autistic children with hyperactivity to act without thinking, to answer even before the questions are finished, not to wait their turn. This suggests, as already stated in the literature (Mansour et al., 2017), that the comorbidity of ADHD may exacerbate cognitive problems on individual with ASD and, thus bring on a greater variety and intensity of deficit than ASD-only. The presence of hyperactivity and behavioral disinhibition in autistic subjects causes deficiencies in the executive functions of working memory, self-regulation, self-control, internalization of speech thus creating problems of control of cognitive activities such as inhibiting inappropriate responses and the difficulty of actively performing the 'goal of direct behavior (Gargaro et al., 2011). Moreover, this study analyses "cognitive flexibility" that is the ability to spontaneously shift to a different action or thought in response to situational changes (Panerai et al., 2014). The results show that there are no differences between ASD+ ADHD and ASD-only groups. In particular, they make the same number of persevered and non-perseverative errors. Therefore, both groups continue to persevere on the same response strategy despite a change of rule. Regarding mental flexibility, this study underlines that group with ASD-only and group ASD+ADHD shown a percentage of perseverative response and errors more highs than ADHD-only children, probably because they implement ritualistic and stereotyped behaviors.

There are significant differences in the number of perseverative errors and total errors between ASD+ADHD group and TD group. The latter are significantly higher in ASD+ADHD group than in the TD group. Furthermore, there is a noticeable difference between ADHD-only, ASD-only, ASD+ADHD groups. In fact, autistic children with and without hyperactivity make more perseverative errors than children with ADHD. The finding of an increase in errors and in particular an increase in perseverative errors among participants with autism is consistent with previous research (Landry & Al-Taie, 2016; Bennetto & Pennington, 1996). In fact, a typical tendency of autistic children is to adopt a classification criterion without being able to abandon it throughout the test, persevering in the first rule identified and maintaining it during the test. Furthermore, non-perseverative errors appear to be absent in autistics with and without hyperactivity, and rarely present in the TD group. Children with ADHD-only make far fewer errors than children with ASD-only but significantly higher than TD children. In conclusion, this study confirmed the difficulty in ASD-only children in executive functioning, due to the presence of ritual and stereotyped behaviors. The central point of the research conducted was to analyze that the presence of hyperactivity in children with ASD worsens their ability to plan and to be flexible at a cognitive level. For this reason, it is very important in clinical practice to analyze not only how executive dysfunctions find expression in ADHD and ASD, but also how such executive dysfunctions manifest themselves when both disorders coexist in the child. Clinical practice shouldn't underestimate comorbidity but understand it and organize a correct intervention. Understanding differences in EF between children with ASD-only, ADHD-only, and ASD+

ADHD is critical to designing individualized intervention strategies. Therefore, the future treatment plan should address the specific cognitive challenges associated with each profile. For this reason, recognizing the impact of comorbid conditions on EF should be useful for long-term planning.

Limit of the research and future prospective

In the present research, there are some limitations. First of all, the small number of participants strongly limits the generalizability of the results, and for this reason future research could aim to extend the sample taken into consideration. To this end, it would be interesting to include in future research, not only the samples already considered in this study, but to understand if there are any differences between the samples examined and a sample of low-functioning autistic subjects. Another important limitation of the present study is the impossibility of using parametric statistics, therefore future studies should increase the sample in order to allow the use of parametric statistics. Further studies should then consider any gender differences. Furthermore, it could be interesting for future research to manipulate the rules of the MCST, using two conditions: an explicit condition, in which the rule to be followed is explained clearly and explicitly, and an implicit condition in which one simply states "yes, right or not, wrong" during the course of the test. This manipulation of the rules will be useful for future research to understand whether or not there are differences between groups.

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Authors' contribution

NA and NF assisted with concept, study design, data analysis, manuscript preparation and manuscript editing; CC and NA assisted with the generation of the initial draft of the whole manuscript, manuscript editing and data interpretation; OS and MAV assisted with manuscript editing and study concept; FG assisted with manuscript editing, data analysis, data interpretation, and study supervision. All authors contributed to and have approved the final manuscript.

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