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Original article

The relationship between theory of mind, executive functioning, and repetitive behavior in high functioning autism spectrum disorder

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SUMMARY

Autism Spectrum Disorder (ASD) is a neurodevelopmental disorder characterised by social communication impairments, and restricted and repetitive behaviors (RRB). Difficulties in Theory of Mind (ToM) abilities in ASD as well as deficits in Executive Functions (EF) are reported in the literature. To date, few studies have investigated the influence of deficits in ToM and EF on ASD symptom severity, including RRB. Thus, the aim of this clinical study is to investigate the relationship between EF, ToM, and RRB in ASD. Our sample consisted of 50 ASD participants divided in two groups: 34 adolescents (mean age = 15.65; SD = 1.68) and 16 adults (mean age = 23.19; SD = 10.08). A mediation analysis evaluated whether the relationship between ToM and EF can be mediated by RRB for each group. Our results demonstrate that RRB plays a key role in the promote the correct relationship between ToM and EF. During development, the structuring of rehabilitation interventions focusing on these aspects can improve abilities to achieve important goals that promote independence and quality of life.

Key words: Autism Spectrum Disorder, executive functions, restricted and repetitive behaviors, theory of mind

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Introduction

Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterised by social communication impairments, restricted interests, and repetitive behaviors [1]. According to the literature, more than half of individuals with ASD do not have intellectual disabilities [2,3] and, through appropriate resources and support, could access the same educational, employment, and social opportunities as their neurotypical peers [4,5]. However, complete independence and integration into the community often remains a challenge for individuals with high functioning ASD, especially in the transition phase from adolescence to adulthood [6,7].

Theory of Mind (ToM) is the social-cognitive ability to understand and infer intentions, beliefs, thoughts, and feelings of others, and to predict their behaviour [8-14]. Several studies indicate that individuals with ASD may have extensive impairment in executive functioning [15-21], especially planning, goal directed behavior, cognitive flexibility and problem solving.

The relationship between ToM and executive dysfunction is still debated, and several studies have attempted to identify the direction of the relationship between the two abilities. Some research suggested that the development of executive function enables the development of ToM [22-27], whereas other authors have argued that ToM abilities are necessary for development of executive control [28-34]. Several longitudinal studies suggest that early ToM (or its precursors) can also predict later executive functions [35-38]. Even if the nature of this relation is still unclear and ambigu-

ous, the evidence currently available seems to indicate a bidirectional relationship between these processes, where some underlying mechanisms are shared [38].
"Non-social" features of autism include RRB, rigidity, and perseverance that, compared with "social" features, are less well understood and studied. Some studies hypothesize that RRB may be associated with impaired executive processes [20,39-42] but the causal relationship between these aspects is largely unknown [43]. For example, it has been suggested that the tendency to persevere on tests involving executive functions may be influenced by circumscribed interests, motor movement patterns, or persistent repetition of activities (such as a rigid routine) or speech that are difficult to interrupt [44,45]. Some evidence suggested that restricted and repetitive behavior are associated with aberrant temporal dynamics of cognitive control brain circuits [46]. A meta-analysis confirmed the relation between executive functions and RRB, emphasising the importance of understanding these relationships for future purposes [43]. RRB in ASD have been shown to be related to deficits in cognitive flexibility, which is the ability to adapt cognitive processing strategies according to environmental demands [17,26,47]. Happé and Frith [18] suggested that executive functioning encompasses the processing of information in context for global meaning, i.e., central coherence. According to the weak central coherence hypothesis, people with ASD tend to be overly focused on details at the expense of understanding the overall meaning of the nature of a specific context [17,18,48]. Fabio et al. [17] demonstrated that cognitive flexibility correlates with a specific social cognition skill, namely facial expression recognition, supporting the hypothesis of an executive deficit in ASD. Furthermore, the authors emphasize that investigating cognitive flexibility in the population with ASD has important clinical implications in terms of understanding RRB.
Limited research attempted to investigate the explanatory power that deficits in ToM and executive functions with respect to changes in ASD symptom severity, including repetitive and stereotyped behaviors [23,42,49].

Objective of the study

Based on these assumptions we designed and conducted a clinical study to investigate the relationship between planning (i.e., executive function), understanding of others' intentions (i.e. ToM), and RRB in ASD. Indeed, understanding the relationship between these variables may provide important insights from a clinical and rehabilitation perspective.

Methods

Participants

Participants were considered eligible for the study according to the following criteria: a diagnosis of Level-1 ASD (high functioning autism) made by a qualified expert in according to the criteria reported by Diagnostic and Statistical Manual of Mental Disorders 5th edition[1] and confirmed using the Autism Diagnostic Observation Schedule - Second Edition [50] and the Autism Diagnostic Interview-Revised [51]; age 11 years and older; absence of other neurological disorders; and absence of pharmacological treatment. Participants were recruited by the Reference Regional Centre for Autism (CRRA) in L'Aquila, Italy. The total sample included 50 participants divided into adolescents (age range between 11 and 18 years old) and adults (more than 18 years old); thus, we obtained a group of 34 adolescents (28 males and 6 females; mean age = 15.65 years, SD = 1.68; mean education duration 9.8 years, SD = 1.77) and a group of 16 adults with ASD (14 males and 2 females; mean age = 23.19 years, SD = 10.08; mean education duration 13.9 years, SD = 1.53). Participants were tested individually in a quiet room. The experimental protocol was approved prior to the recruitment of participants by the Ethical Committee of the NHS Local Health Unit. Participants or tutor-by-law provided their informed consent to participate in the study after being properly briefed about the aims of the study and the confidentiality of data processing. The study followed the principles established by the Declaration of Helsinki.

Measures

Clinical measures

The Autism Diagnostic Interview-Revised [51,52] is a semi-structured clinical interview conducted with a caregiver, based on the International Classification of Diseases (ICD-10) [53] and focused on ASD symptoms such as abnormal reciprocal interaction, abnormal communication, and restricted and repetitive behavior. This interview is composed of 93 ordinal-scaled items where only part of them is used in the algorithm for autism classification. ADI-R is composed of five sections regarding concerns, communication, social development and play, repetitive and restricted behaviors, and behavioral problems.
The Autism Diagnostic Observation Schedule-2 Edition [50] represents a semi-structured assessment of communication, social interaction, restricted and repetitive behaviors; the activities provided are directed at stimulating all those behaviors significant to the diagnosis of ASD. The ADOS-2 consists of 5 different modules, selected in relation to language skills and age. According to age and maturity level of participants, we used Module 3 or Module 4, both designed for verbally fluent individuals.

ToM measure

The Intentions Attribution - Comic Strip Test [54] (IAT) is a

type of mentalizing nonverbal test. It consists of a series of cartoon-type stimuli to assess the ability to attribute and understand other's intentions. Participants are shown a sequence of three vignettes in which there is a character performing an action and then they are asked to identify one of the three alternative proposed endings that represent the intention behind the action. This test aims to assess the ability of an individual to deduce the character's intention. The test is divided into two conditions: 1. First Order Intention Attribution (14 items) and 2. Second Order Intention Attribution (12 items). There is a Control Condition (12 items) describing physical causality with human figures and objects. Each subject can get a score between 0 and 1 for each story. To our knowledge, to date, there are no standardized tests that allow assessment of nonverbal attribution of intentions.

Executive function measure

The Tower of London (ToL) was originally developed by Shallice [55]. ToL is considered a measure of planning abilities and a poor performance during this task is interpreted as an inefficient performance in planning processes [55,56]. The instrument is composed by a wooden board with three pegs of different length with three different coloured balls inserted on them. Starting from an initial disposition of the balls, participants are asked to move them to match a goal disposition. The average time to reach the goal disposition and number of moves are usually measures considered to assess the performance during the task. ToL is a tool that assesses higher order problem-solving abilities and can identify deficits in the planning processes involved in generating a plan to address new demands.

Statistical analysis

Regression analysis

We performed a linear regression analysis. Variables for regression analysis included measures of intention's attribution (AIT), RRB (reported by ADOS-2 or ADI-R) and planning (ToL). Specifically, for the adolescent group we considered the total score of IAT, the total score of ToL and RRB derived from ADI-R. For the adult group, we considered the total score obtained on the second-order intention attribution items of the IAT because it requires more difficulty in the process of inferring character intention. This decision is supported by evidence that ToM continues to develop throughout childhood, adolescence, and adulthood [57-63]. ToM ability improves with age [64] so items in the first-order series might be too simple and easily completed by an adult individual. Regarding planning skills, we divided the ToL test according to the number of moves required to move from start-to end-state and considered only the average of the scores obtained at the "difficult" trials, namely those requiring 5 moves. This approach is consistent with the hypothesis that increasing age would lead to better performance on the more difficult tests of the ToL. According to Albert and Steinberg [65], although mature performance on ToL was attained by age 17 on relatively easy problems, performance on the hardest problems showed improvements into the early 20s. Furthermore, as pointed out by Hill [19], planning difficulties in individuals with autism are evident at the most complex levels [66], which would correspond to the planning required in everyday life. Finally, we decided to consider RRB derived from the ADOS-2 Module 4 because the ADI-R may not be reliable in adults without intellectual disability [67]. In addition, parents of younger individuals with ASD may be more sensitive to the reporting of RRB, whereas parents of older individuals may be habituated to these behaviors and thus less likely to report them. In addition, it is highly likely that the frequency and characteristics of RRB change with age [68-72].

Mediation analysis

Mediation analysis allows to understand the mechanism through which the causal variable effects the outcome [11,73,74]. In mediation process the association between the independent variable X and the dependent variable Y involve one (or more) mediator variable (M) that is able to explain the relation cause-effect between X and Y [75]. In order to explore the significance of mediation effect we used the Sobel test [76,77]. Specifically, the Sobel test was performed to evaluate whether the constructs measured by ToL and IAT were mediated by RRB, as one of characteristic symptoms of ASD in both adolescent and adult groups. The Sobel test involves computing the ratio of ab to its estimated standard error [11]. The results were considered significant at a threshold of $p < 0.05$. The analysis was performed using The Statistical Package for the Social Sciences (SPSS) software.

Results

Two regression analyses were conducted for each age group separately: one with executive function as the dependent variable and intention's attribution as the independent variable, and another one with executive function as the dependent variable and RRB as the independent variable. Based on the results, a mediation analysis was run with intention's attribution as the independent variable, executive function as the dependent variable and RRB as the mediator.

Adolescent group

We conducted a first regression analysis using the planning ability as the dependent variable and the intention's attribution as the independent variable ($R2 = 0.41$,

TABLE I. *Results of regressions analysis.*

Group	Model	Variables	b (SE)	*P*
Adolescents	1	Planning abilities (DV) Intention attribution (IV)	0.17 (0.05)	0.002
	2	Planning abilities (DV)	-0.32 (0.12)	0.02
		Restricted and repetitive behaviors (IV)		
Adults	1	Planning Abilities (DV) Intention Attribution (IV)	0.49 (0.19)	0.02
	2	Planning abilities (DV) Restricted and repetitive behaviors (IV)	-1.10 (0.36)	0.01

b = 0.17, SE = 0.05, p = 0.002). This was followed by a second regression analysis where RRB, measured by ADI-R, became the predictor, while ToL was the dependent variable (R2 = 0.32, b = -0.32, SE = 0.12, p = 0.02). We run a mediation model based on regression results (Tab. I). The relationship between Intention Attribution (X) and planning (Y), using the ADI-R RRB items as mediator, was explored. The Sobel test showed that this model was significant (SE = 0.02; p = 0.03).

Adult group

We conducted a first regression analysis using the planning ability as the dependent variable and the intention's attribution as the independent variable (R2 = 0.32, b = 0.49, SE = 0.19, p = 0.02). This was followed by a second regression analysis where RRB, measured by ADOS-2, module 4 became the predictor, while planning was the dependent variable (R2 = 0.47, b = -1.10, SE = 0.36, p = 0.01). We run a mediation model on the basis of the regression results. The relationship between Intention Attribution (X) and planning (Y), using the ADOS-2 RRB items as mediator, was explored. The Sobel test showed that this model was significant (SE = 0.27; p = 0.04).

Discussion

The present study examined the role of the RRB on two skills, namely planning as ability of executive functioning and intention's attribution as a ToM ability, in a sample of adolescents and adults with high functioning ASD. Mediation analysis results showed that in both groups the ability to understand and attribute others' intentions (independent variable) and the planning ability (dependent variable) were mediated by a symptomatic characteristic, the RRB (M). Specifically, the observed effects suggest that RRB negatively influences the relationships between intention's attribution and planning ability.

Our models support the hypothesis that the ability to represent one's own mental states and those of others is necessary to "strategically" control thoughts and behaviors [78-80]. The ability to infer the intentions of others is achieved very early in development [10,81,82], whereas proper maturation of executive functions appears to occur during adolescence [83]. Similarly, RRB emerge early in ASD [84-86]. As explained by Wade and collaborators [38], tasks that require the ability to plan and inhibitory control need the awareness that actions to achieve the goal are first mentally represented. Thus, the ability to exercise the executive control depends on the ability to understand that mental states are causally related to actions and behaviors. Our results showed that in individuals with ASD this relationship could be disrupted by the presence of RRB. Indeed, better understanding of others' intentions and fewer RRB should indicate better planning skills. Identifying the relationship between the characteristics of ASD that represent a barrier to develop abilities is critical to structuring, as early as childhood, interventions to improve impaired skills and support the individual toward achieving developmental milestones, thereby improving quality of life [87,88]. It would be useful to structure interventions to improve ToM and social skills, as well as offset the impact of RRB on daily functioning, to promote executive skills that develop later.

Despite the interesting results, our study has limitations. A potential limitation concerns the sample size, a critical methodological item concerning the distribution of the product term; however, our sample size ensures the conditions for analysis.

Future studies should test the mediation model considering gender as a possible interaction variable. In addition, future studies should consider a neurotypical developmental control group. Executive functions is an umbrella-term that comprehends a series of processes [89] but in our study we considered only planning skill.

In conclusion, our results indicate that RRB plays a key role in promoting the correct relationship between ToM and planning. Working on these aspects during the development of ASD children and adolescents appears a logic strategy to improve abilities and achieve important goals toward independence and quality of life.

Conflict of interest statement

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest

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Authors' contributions

MV and MM, designed the research and the data analysis; MA analysed data; ILD and AB collected the data; all the authors contributed in writing the paper.

Ethical consideration

The experimental protocol was approved prior to the recruitment of participants by the Ethical Committee of the National Health System Local Health Unit (Comitato Etico delle Province di L'Aquila e Teramo, approval reference number 0052505/21).

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