Behavioral Profile of Executive Dysfunction in Children and Adolescents with Autism Spectrum Disorder

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Abstract

Background: Executive dysfunction has been proposed as a fundamental impairment in children with autism spectrum disorder (ASD), however, existing findings are inconsistent.

Objectives: The present study aimed at evaluating the behavioral profile for executive functions (EF) in adolescents compared to healthy individuals using the Farsi translation of the behavior rating inventory of executive function (BRIEF).

Methods: In this study, 34 participants (aged 5-16) with ASD were compared to 36 age and gender matched typically developing (TD) children using the BRIEF and childhood autism rating scale (CARS), and raven progressive matrices (RPM).

Results: All subscales of the BRIEF were significantly higher in children and adolescents with ASD. Working memory was impaired in 88% of the ASD group, and the inhibition subscale had the highest mean score. The visual response subscale of CARS correlated significantly with the metacognition index (MCI) and global executive composite (GEC) of the BRIEF. A slight negative significant correlation was found between the BRIEF planning/organization T-score and age in the group with ASD.

Conclusions: Adolescents with ASD have difficulties in every day executive functioning, mostly in working memory and inhibition. These deficits are related to some aspects of social and sensory impairments seen in ASD.

Keywords: Autism Spectrum Disorder, Behavioral, Test, Executive Functions

1. Background

Executive function (EF) is a collection of mental control processes necessary to sustain effective goal-directed behavior (1). EF is essential to facilitate physical, cognitive, and emotional self-control (2-4) and begins to develop during the first years of life (1, 4). EF indices include response inhibition, working memory, cognitive flexibility, fluency, and planning (4, 5). EF plays an important role in children's cognitive functioning, behavior, emotional control, and social interactions (6, 7), and its deficits have been repeatedly observed in neurodevelopmental disorders (8).

Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterized by deficits in social interaction, communication, and markedly restricted, repetitive patterns of behavior, interests, and activities (9). Deficits in EF have been reported in ASD (4, 10-12) and proposed as

a causal factor for its stereotyped behaviors (13). Executive dysfunction may also account for social difficulties and behavioral problems, as it is linked to theory of mind (TOM) (13-15), which refers to the ability to attribute mental states to others (14, 16, 17). For example, some authors found a relationship between working memory and inhibition with TOM in children with ASD (13).

There are various findings on executive dysfunction in children with ASD. These children display perseverative responses in the Wisconsin card sorting test (WCST), which is one of the frequently used measure of EF (18, 19). There is clear evidence for shifting impairment as a main component of EF, however, literature on other components of EF impairment in ASD is inconsistent (20, 21). Complexity of the construct of EF may make it difficult to establish specificity of deficits and behavioral constellations (22). Nevertheless, controversy in findings may be related to certain

measures used and patient characteristics such as age and IQ, and co-occurring disorders (20, 23).

Each neuropsychological test measures a narrow cognitive function in a laboratory situation (24), which may be different from everyday functioning. Behavior rating inventory of executive function (BRIEF) (25) is a behavioral measure designed to assess executive functioning in real life including behavioral and emotional regulation. BRIEF studies have consistently shown a higher impairment of shifting in ASD (26-31).

Considering the existing controversies on executive dysfunction findings in ASD and limited related studies in Iranian children, we aimed at assessing the real world executive control in a group of Iranian children and adolescents with ASD compared to a typically developing group.

2. Objectives

This study aimed at assessing the behavioral profile of executive function in children and adolescents with ASD compared to a normal group using a parent-rated questionnaire.

3. Materials and Methods

3.1. Participants

Participants were 5 to 16 year old adolescents (no = 34) with confirmed clinical diagnosis of autistic disorders by board-certified child and adolescent psychiatrists, recruited either from a referral child and adolescent psychiatry clinic (n = 14) or schools for children with special needs (n = 20). The comparison group included typically developing (TD) children (no = 36) recruited from mainstream schools. Exclusion criteria were any serious medical or neurological disorders, or an IQ below 70.

3.2. Measures Behavior Rating Inventory of Executive function (BRIEF) - Parent Form

This scale, which consists of 86 items, was designed to measure components of EF in children aged 5 to 18. Parents rate their child's behaviors on a 3-point liker scale (never, sometimes, and often). The measure comprises 8 empirically derived subscales. The following subscales sum up to the metacognition index (MCI): inhibit, working memory, plan/organize, organization of materials, and monitor. Shift and emotional control subscales contribute to the behavioral regulation index (BRI). MCI and BRI further combine into an overall construct defined as the Global Executive Composite (GEC) (32). The process of translation to Farsi and back translation of the BRIEF had been done previously and used in a group of Iranian children with attention deficit hyperactivity disorder (33).

3.3. Childhood Autism Rating Scale (CARS)

The CARS is a 15- item scale for rating social, emotional, and sensory symptoms of autism (34). A trained examiner rates the symptoms based on parents report and observation of the child. Each item is scored from 1 (no abnormality) to 4 (severe abnormality). The score 30 is the cutoff point for the diagnosis of autistic disorder.

3.4. Strengths and Difficulties Questionnaire (SDQ)-Parent Form

This 25-item scale (35) has 5 indices (ie, hyperactivity, emotional problems, conduct problems, peer problems, and prosocial behavior) and a total difficulties score. The SDQ has been validated for Iranian children (36) and adolescents (37).

3.5. Raven's Progressive Matrices (RPM)

RPM (38) is a nonverbal measure for intelligence and perceptual reasoning. This test has been standardized for Iranian children and adolescents aged 5 to 18 years (39).

3.6. Procedure the ASD Group

All children and adolescents were clinically diagnosed as having autistic disorder, Asperger disorder, or PDD-NOS based on the diagnostic and statistical manual of mental disorders, fourth edition (DSM-IV-TR) (40) criteria. There were some symptoms of inattention and hyperactivity-impulsivity in the participants. However, as the comorbidity of ADHD and ASD is not allowed based on the DSM-IV-TR, we ruled out ADHD diagnosis in the participants; these symptoms were better accounted for ASD. Parents were asked to complete BRIEF, based on the children's behavior in the last 6 months. They also completed a questionnaire in which children's demographic characteristics, developmental milestones, list of current prescribed medications, age at diagnosis of ASD, and parents' level of education were inquired. The CARS was used to evaluate the severity of autistic symptoms. Raven's Progressive Matrices was used to provide an estimation of intellectual ability. We did not change any therapeutic interventions being used for the participants during the study.

3.7. The Typically Developing (TD) Control Group

The parents of the control group were asked to complete the demographic questionnaire, and parent forms of the BRIEF and SDQ. With respect to the Iranian normative studies, children with SDQ total score above 15 were excluded. The intelligence abilities were measured using the Raven's progressive matrices.

3.8. Statistical Analysis

Using the SPSS version 16.0, group matching was evaluated by independent t test, Mann–Whitney U test, and chi-square. Analysis of covariance (ANCOVA) was used to explore the effect of ASD vs. TD for executive impairments after controlling the non-verbal IQ.

3.9. Ethical Considerations

This study was approved by the ethical committee of Tehran University of Medical Sciences. The objectives and procedure were explained to the parents, and written informed consent was obtained.

4. Results

4.1. Participants' Characteristics

Table 1 demonstrates the participants' demographic characteristics. There were no significant differences between the 2 groups in age and gender. However, the IQ of the TD group was significantly higher than the ASD group. The mean age of the diagnosis of ASD was approximately 6 years (SD = 2.5 years).

Based on the DSM-IV criteria, 23 adolescents in the ASD group were diagnosed as having an autistic disorder, 2 as Asperger disorder, and 9 as PDD NOS. Their mean CARS total score was 36.74 (SD = 6.5), with min = 25 and max = 55.5. Nearly 80% of the ASD group was taking medications, mostly risperidone (64%) and Ritalin (32%), respectively. of them, 12 did not receive any medications.

The mean SDQ total score in the TD group was 9.8 (SD = 4.19), with min = 1 and max = 15.

4.1. Group Differences Based on BRIEF Scores

Table 2 demonstrates group related differences of executive performance. A large effect (0.50) of ASD on global executive composite was revealed by ANCOVA after controlling the nonverbal IQ [$F (1, 66) = 66.9$, $P < 0.001$]. Besides, a main effect of ASD on executive dysfunction was found in all subscales of BRIEF ($P < 0.001$). The partial eta squared statistic of working memory (0.46), inhibit (0.45), and plan/organize (0.42) subscales indicated that their effect sizes were 'large'. The domain of organization of materials had the least effect size (0.20).

The frequency of those with ASD, who had impaired working memory was highest compared with other EF scales considering the T- scores (82.4%), followed by initiate (76.5%), inhibit (70.6%), and monitor (70.6%) subscales. This frequency had the least amount in organization of materials subscale (20.6%).

With regards to association between age and BRIEF scores, there was no significant correlation between the raw scores of BRIEF and age in the 2 groups. However, a significant negative ($P < 0.05$) correlation was found between the planning/organization T- score and age in the group with ASD.

4.2. Correlations Between BRIEF and CARS in the ASD Group

Table 3 demonstrates the association among subscales of these 2 measures. Visual response subscale of CARS significantly correlated with the Metacognition Index (= 0.279, P = 0.036) and global executive composite (= 0.267, P = 0.043) of the BRIEF.

5. Discussion

The purpose of the present study was to explore executive dysfunction in children and adolescents with ASD in comparison with a typically developing group using the BRIEF parent form, a questionnaire which measures executive control of daily activities.

The findings suggest that EF is impaired in children with ASD. Inhibition and working memory were the most impaired components in the group with ASD. The percentages of participants, whose score were above the clinical cutoff were between 20.6% (organization of materials = OM) and 82.47% (working memory = WM). The next highest percentages in our participants with ASD belonged to initiate, inhibit, planning, and shift subscales (76.5, 70.6, 66.7 and, 61.8, respectively). In Van den Bergh study, these frequencies were from 20 (for domain of planning) to 50 (for domain of cognitive flexibility). Kenworthy et al. (2005) reported these percentages for a group of 72 individuals aged 5 to 17 years, who were diagnosed as having high-functioning autism (HFA) or Asperger disorder (AD) including inhibit, shift, P/O, and WM (64 for HFA and 38 for AD). They confirmed flexibility and organization as the most impairments found.

There are mixed results in the literature in inhibitory control of individuals with ASD (41). Inhibition, shift, and working memory are 3 fundamental components of EF and are interrelated (42). Some evidence shows that there is substantial need of inhibition for working memory processes and they are both necessary for shifting (6, 43). Impairments of inhibition may cause intrusion errors in children with ASD (They may fail to suppress irrelevant thoughts) (44).

Blijd-Hoogewys (2014) found that the shift subscale was clinically high and showed the cognitive disability of the ASD group. The inconsistent findings of previous studies may be due to developmental variations and compensatory mechanisms in children with ASD (6, 45). Some studies investigating working memory in ASD (5, 46) independent of inhibitory skills showed no WM deficit.

Table 1. Demographic Characteristics of the ASD and TD Groups

Characteristic	ASD (n = 34)	TD (n = 36)	P
Mean age (months)	117.35 (35.52) Min = 60, Max = 192	116.9 (34.16) Min = 60, Max = 180	0.957
Gender	Male 29 (85.29%)	Male 29 (80.55%)	0.754
Non-verbal IQ	80.82 (10.74) Min = 70, Max = 107	110.03 (13.53) Min = 81, Max = 131	< 0.001

Table 2. Group Related Differences of the BRIEF Scores

	T Score		F-testb			% Impaired in the ASD Group
	ASD (n = 34)	TD (n = 36)	F (1.67)	P	$N_p{}^2$	T Scores $\geq$ 65
Inhibit	71.76 (12.89)	46.75 (6.08)	53.97	< 0.001	0.45	70.6%
Shift	69.03 (12.14)	52.11 (10.50)	18.29	< 0.001	0.21	61.8%
EC	66.12 (10.40)	52.03 (8.95)	23.03	< 0.001	0.26	55.9%
Initiate	67.41 (19.23)	51.89 (7.81)	33.92	< 0.001	0.34	76.5%
WM	70.27 (8.98)	50.19 (7.86)	55.93	< 0.001	0.46	82.47%
PO	68.03 (9.77)	50.81 (7.72)	47.54	< 0.001	0.42	66.7%
OM	57.03 (10.38)	47.17 (9.13)	16.78	< 0.001	0.20	20.6%
Monitor	67.53 (9.78)	47.53 (8.78)	43.19	< 0.001	0.39	70.6%

Abbreviations: EC, emotional control; OM, organization of materials; PO, plan/organize; WM, working memory.

Table 3. Correlations Between the BRIEF and CARS Subscales in the ASD Group

BRIEF	Relationship to Others	Imitation	Emotional Response	Body Use	Object Use	Adaptation to Change	Visual Response	Listening Response	Taste-Smell Touch Response and Use	Fear and Nervousness	Verbal Communication	Nonverbal Communication	Activity Level	Level and Consistency of Intellectual Response	General Impression
Total scores	0.086	0.119	-0.072	0.199	-0.039	0.018	0.202	0.181	0.016	0.204	0.108				
Inhibit	0.068	0.120	0.129	0.038	0.148	0.253	0.168	0.038	-0.119	-0.109	0.072	0.082	0.135	-0.131	0.053
Shift	0.199	0.297[a]	0.120	0.002	-0.016	-0.004	0.316[a]	0.091	-0.267[a]	-0.115	0.095	-0.020	-0.098	-0.064	0.183
EC	0.080	0.150	0.052	-0.062	-0.030	0.040	0.065	0.182	-0.137	-0.127	-0.018	-0.162	-0.176	-0.102	0.072
Initiate	0.318[a]	0.106	-0.022	-0.120	0.073	0.271[a]	0.368[b]	-0.018	-0.077	-0.040	0.004	0.138	0.167	0.052	-0.030
WM	-0.025	0.077	0.038	0.066	0.069	0.129	0.070	0.010	-0.261[a]	-0.028	-0.096	-0.142	0.020	-0.262	-0.138
PO	0.172	0.215	0.059	-0.179	-0.017	0.190	0.030	-0.047	-0.013	-0.188	0.087	-0.106	0.034	-0.222	-0.166
OM	0.239	0.164	0.152	0.133	0.070	0.028	0.281[a]	-0.040	0.000	-0.083	0.055	0.107	0.250	-0.046	0.107
Monitor	0.124	0.316[a]	0.210	0.232	0.126	0.244	0.298[a]	0.070	-0.148	0.008	0.032	0.064	0.040	-0.080	0.073

Abbreviations: EC, emotional control; WM, working memory; PO, plan/ organize; OM, organization of materials.
[a] P< 0.05.
[b] P< 0.01.

There is a possibility that people with ASD, who are recruited from a referral psychiatric center, have greater cognitive impairment as well as the presence of comorbid disorders, which might over-represent cognitive disorders. Therefore, inhibitory control may influence several cognitive skills and may be a cause of the predominance of inhibition and working memory dysfunction in this ASD group.

In this study, positive significant correlations were found between the severity of some social symptoms of ASD and executive dysfunction. The association found between the initiate index with relation to others and adaptation to change can support the role of executive dysfunction in social interactions problems among ASD individuals. Moreover, the highest correlation found was between the visual response index of CARS and the MCI as well as shift, initiate, organization of materials, monitor, and the global executive composite (GEC). These findings were consistent with those of Kenworthy (2009) who found significant relationships between both laboratory tasks and behavior rating scales of executive functions and autism symptoms.

Magyar and Pandolfi (47) found that visual response accompanied with verbal communication, nonverbal com-

munication, relating to people, and imitation items of CARS, load into a construct regarded as social features of ASD. Keehn et al. (48) discovered that the function of a visuospatial orienting network is impaired in ASDs. This network is a component of attention that transfers selected visual inputs towards an executive control network. On the other hand, verbal and nonverbal communications were the only social items that did not correlate significantly with BRIEF items in our study. Although it has been proposed that inner speech abilities can help EF skills, Holland and Low (49) showed that in ASDs unlike TDs, inner speech played no important role in an arithmetic task-switching executive control test and that these people were dependent on visuo-spatial resources for completing that task. Joseph et al. (13) claimed that ASDs use less language skills in EF control. They did not find any specific relationship between EF and verbal abilities in their ASD group.

The item taste/smell/touch response and use of CARS had a significant negative correlation with working memory and shift components of EF. A possible explanation for this result may be that modified sensory functions of ASDs act as compensatory mechanisms for executive dysfunctions. Baron-Cohen et al. (50) proposed that sensory hypersensitivity justifies talent in ASDs at the sensory level. Sensory hypersensitivity can provide excellent attention to details (in perception and memory) and results in strong local information processing that can cause a faster analysis of the whole. However, Minshew and Hobson (51) described reports of ASDs who experienced sensory overstimulation as overwhelming and disabling. Also, Happe (52) indicated perceptual features of ASD may underlie the autistic need for sameness. Even though limited work has been done on mechanisms and consequences of sensory abnormalities in ASDs, sensory hypersensitivity theory may explain how sensory symptoms are associated with a higher EF performance in this study.

There is some evidence supporting the association between age and EF through normal development. Based on the results of the BRIEF in normal children, Huizinga and Smidts (2011) (53) reported a decrease in executive dysfunction with increasing age. Anderson (2010) (54) suggested that different EF components emerge through various stages of life. Regarding age-related differences in executive function in children with ASD, a study administered BRIEF to a group of 6 to 18 year olds diagnosed as having ASD (54). The highest score was related to the inhibit subscale in 6 to 8 year olds; however, the greatest score belonged to planning in 12 - 14 year olds in comparison with 10 to 12 year olds. Rosenthal et al. (2013) (55) in a cross-sectional cohort of 185 children with ASD found that the initiate, working memory, and organization of materials subscales scores worsened significantly with increase in

age. The difference on metacognitive executive abilities between the ASD and healthy group increased as they grew older. Our study revealed a slight negative significant correlation between planning/organization T- score and age in the ASD group. This was not observed in the TDs' raw or standardized scores or even in other EF domains.

5.1. Limitation

This study had some limitations. The sample size was small and the age range was broad, the facts which can limit the generalizability of our results. Moreover, Parents' reports were the only measures being used to evaluate the executive dysfunctions in the participants. Moreover, our participants were recruited from a referral center and could suffer from comorbid disorders and their greater cognitive impairment.

5.2. Conclusion

In summary, this study suggests that the adolescents with ASD have difficulty in executive function, mostly working memory and inhibition, which can be reflected in their real world experiences. In this study, we did not use any research tool to evaluate comorbid psychiatric disorders except for psychiatric assessment. These problems are associated with some features of social and sensory impairments seen in ASD.

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Footnotes

Authors' Contribution: Hoda Mahdavi designed the proposal of study, collected all data, performed statistical analysis and provided primary draft of the manuscript. Zahra Shahrivar conceived and designed the study, interpreted the data and revisited the manuscript. Mehdi Tehrani Doost helped design the study, re-analyzed statistical data, and revised the manuscript. Yasaman Fatholahi and Azraa Jahanitabesh administered the IQ test and CARS. Alia Shakiba helped edit the draft of the manuscript. All authors read and approved the final manuscript.

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