Impaired executive functions predict repetitive behaviors in toddlers under 36 months old with autism spectrum disorder symptoms

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Abstract

Background: Executive Functions (EFs) deficit in Autism spectrum disorder (ASD) has been mainly investigated, while less is known about the EFs in toddlers. The study aimed was to investigate the relationship between EFs deficits and repetitive and restricted behaviors (RRBs) in toddlers with ASD symptoms. Finally, we examined whether EFs deficits were predictive of RRBs.

Method: Cross-sectional data were collected from mothers of forty-five toddlers under 36 months old. The modified checklist for autism in toddlers (M-CHAT), the Gilliam autism rating scale (GARS-2), the behavior rating inventory of executive functioning-preschool version (BRIEF-P), and repetitive behavior scale-revised (RBS-R) administered to mothers.

Findings: We found significant associations between parent-reported executive functions problems and (1) stereotyped behaviors, (2) self-injurious behaviors, (3) ritualistic behaviors, (4) sameness behaviors, (5) restricted behaviors, (6) compulsive behaviors, and (7) repetitive behaviors total score. There was a lack of association between M-CHAT and social interactions deficits of GARS-2 and the repetitive behaviors total score. Increases in the degree of EFs deficits predicted increments in repetitive behaviors.

Discussion: These results support a link between executive dysfunction and RRBs. Future research on RRBs in ASD may benefit from focusing on specific executive functioning abilities rather than general categories.

Introduction

ASD is a neurodevelopmental disorder characterized by deficits in social interaction and social communication and a range of restricted, repetitive, and stereotyped patterns of behavior, interests, and activities (American Psychiatric Association, 2013b). The onset of ASD symptoms usually occurs in symptoms of autism in infancy and toddlers before the age of 2 years (Chawarska et al., 2014). RRBs are a hallmark of ASD (American Psychiatric Association, 2013b; Boyd et al., 2012). The RRBs manifest across echolalia, stereotyped behaviors, self-injurious behaviors, ritualistic behaviors, sameness behaviors, restricted behaviors, compulsive behaviors, and hyper- and hyporeactivity to sensory stimuli (South et al., 2005). Usually, RRBs are divided into two categories: higher-order repetitive behaviors (HORBs) and lower-order repetitive behaviors (LORBs). LORBs include motor actions (e.g., self-injurious behaviors, repetitive use of objects, and stereotyped behaviors). In contrast, HORBs include repetitive and restricted cognitive, interests or complex behaviors like compulsive behaviors, routines, and rituals, insisting on sameness and consistencies and restricted interests (Leekam et al., 2011; Szatmari et al., 2006). RRBs have significant impacts on different aspects of psychosocial adjustments of the people with ASD. These symptoms interfere with social interactions, learning new skills, and participating in daily life activities (Shiri et al., 2020).

Although in recent years new theories have been developed to explain different features of ASD core impairments, yet no neuro-psychological theory has been proposed that can satisfactorily explain all of ASD aspects. EFs, theory of mind (ToM), and weak central coherence cognitive theories have dominated neuropsychological research into ASD (Demetriou et al., 2019; Rajendran & Mitchell, 2007). It is assumed that the theory of mind and the theory of weak central coherence could explain many of the deficits in social interaction and social communication associated with ASD (Baron-Cohen et al., 1985). However, the EFs theory may provide the best explanation for the non-social aspects of ASD, such as repetitive behaviors and restricted interests (Damasio & Maurer, 1978; Frith & Happe, 2004; Hughes et al., 1994; Joseph, 1999). EFs are a set of cognitive skills that defined as an “umbrella” term include goal-directed and self-regulatory cognitive abilities such as inhibition, shifting, working memory, flexibility, and planning (Diamond, 2013; Wallace et al., 2016). Additionally, EF is the only theory that acknowledges both the motor and cognitive characteristics of ASD (e.g., perseveration, insistence on sameness, repetitive hand flapping, and rocking) (Rajendran & Mitchell, 2007).

Lopez et al. (2005) investigated the relationship between EFs and the RRBs symptoms of ASD in adults. These researchers reported that working memory, flexibility, and response inhibition were highly related to the restricted, repetitive symptoms. South et al. (2007) showed a significant relationship between EFs and RRBs and no significant correlation between central coherence ability and RRBs in nineteen individuals (ages 10–19) with high-functioning autism. They were reported that set-shifting ability predicted RRBs in people with ASD. Mosconi et al. (2009) found that a deficit in inhibitory control ability is correlated with HORBs in eighteen 15 aged adolescents with ASD. Boyd et al. (2009) showed EFs deficits associated with RRBs in school-aged children with ASD. Previous studies also demonstrated a relationship between RRBs and EFs deficits such as set-shifting (Jones et al., 2018; Miller et al., 2015), inhibition (Jones et al., 2018), and planning (Van Eysen et al., 2015). Recent meta-analyses study by Iversen and Lewis (2020) confirmed significant associations between HORBs and poor EF abilities. Previous studies have examined the relationship between EFs and RRBs in school-age children, adolescents, and adults with autism spectrum disorder using performance-based standardized assessment of EFs. Therefore, we have no information at an early age.

Early research in domains cognitive domains of ASD provides valuable opportunities to study autism before confounding effects of interventions, development of compensatory strategies, and comorbid syndromes and disorders have begun to impact its manifestation. So, the primary goal of this study was to examine the relationship between EFs and the restricted, repetitive behaviors in toddlers with ASD symptoms. Our study set out to initially replicate the EFs using a well-established EFs inventory in toddlers. Based on the EFs of ASD, we predicted that impairments in EFs would be highly correlated with RRBs in toddlers. Similarly, we hypothesized that deficits in EFs would predict a significant portion of the variance in RRBs. While, deficits in EFs would not predict a significant portion of the variance in social interaction skills in toddlers.

Methods

Participants
Participants included forty-five toddlers (34 males, 11 females; mean age = 26.33 months; range 16–36 months) were referred to autism center. Each of the subjects was diagnosed using comprehensive evaluation that included standardized testing, behavioral observation, and extensive parent reports gathered through interviews and questionnaires. Diagnoses were based on DSM 5 guidelines (American Psychiatric Association, 2013a). All subjects were evaluated by an ASD specialist with a Ph.D. in clinical psychology and at least one assistant (with at least a master's degree in clinical psychology). The diagnosis was made based on informed clinical judgment following interaction with the child, formal testing, and review of parent reports and records. The Gilliam autism rating scale—second edition (GARS-2) (Gilliam, 2006) and the modified checklist for autism in toddlers (M-CHAT) (Robins et al., 2014) were also administered to parents of the subjects. To participate in our study, parents completed an informed consent form for the child’s participation, executive functions and feeding problems measures (described below), and a demographic information checklist. Inclusion criteria included no known comorbid psychological and medical disorders, or hearing and visual deficits. All study procedures were performed after the study was approved by the Ethics Committee of the Shahid Beheshti University.

**Study Measures**

Demographic Data: Demographical information was collected by checklist included: age, sex, age of parents, parent’s educational degree attainment, and employment status.

**Modified checklist for autism in toddlers (M-CHAT)**

The M-CHAT (Robins et al., 2001) is a 23-item (yes/no) validated developmental autism screening checklist for toddlers. The M-CHAT administered to parents (Dumont-Mathieu & Fein, 2005). A higher score means a higher risk of autism in the child. In this study, the internal consistency of the M-CHAT was obtained by Cronbach’s alpha for the whole scale of 0.80.

Gilliam Autism Rating Scale (GARS-2): GARS (Gilliam, 2006) is a well-known scale that assists specialists in identifying and diagnosing ASD in individuals and evaluating the severity of the child’s ASD symptoms. GARS-2 includes 42 items and three subscales: stereotyped behaviors, communication, and social interaction. GARS-2 has good sensitivity and specificity in Iranian children (Gorji et al., 2020).

**Behavior rating inventory of executive functioning-preschool version (BRIEF-P)**

BRIEF-P (Gioia et al., 2003) is a standardized rating scale developed to provide a window into everyday behaviors associated with specific domains of executive functioning (EF) in children ages 2 to 5.11 years old. The BRIEF-P includes 63 items in five non-overlapping scales. The EFs in this scale are divided into nine subscales. The five clinical subscales consisted of, shifting, inhibition, emotional control (EC), working memory, and planning /organization. Also, this scale consisted of inhibitory self-control index (ISCI) that is composed of the inhibition and EC subscales, the flexibility index (FI) is composed of the shift and EC subscales, and the emergent metacognition index (EMI) that is composed of the working memory and plan/organize subscales. This scale total’s score makes global executive composite scale (GEC). The validity and reliability of BRIEF-P have been reported in previous works (Ezpeleta et al., 2015; Isquith et al., 2005; Sherman & Brooks, 2010). Sadeghi et al. (2019b) reported that BRIEF internal consistencies for all subscales were high in Iranian children (Cronbach’s = 0.80 until 0.97).

**Repetitive Behavior Scale-Revised (RBS-R)**

This scale (Bodfish et al., 2000) is a 43-item parent rating scale intended to evaluate six dimensions of repetitive behavior (Stereotyped Behavior, Self-Injurious Behavior, Compulsive Behavior, Ritualistic Behavior, Sameness Behavior, and Restricted Behavior). Lam and Aman (2007) showed construct validity for subscales from 0.68 to 0.98 and internal consistency of subscales from 0.78 to 0.91. These part descriptions were mostly reproduced from our previous studies using a similar scale (Sadeghi et al., 2019a; Sadeghi et al., 2021). RBS-R has already been validated into the Iranian population (Khamoushi & Mirmahdi, 2015; Rezayi & Iari lavasani, 2017).

**Statistical Procedure**

The data were summarized with descriptive statistics using SPSS 22 software (Corp, 2013). Correlations between variables were estimated by Pearson correlation. Also, associations between EFs and the feeding problems were estimated by linear regression.

**Results**

Participants in this study included 45 toddlers with ASD symptoms (34 males and 11 females). Participant’s demographical information is included in Tables 1 and 2. Also, descriptive statistics of the study variables are presented in Table 3.

| Variable          | M     | SD    | Min | Max |
|-------------------|-------|-------|-----|-----|
| Age of children   | Month | 26.33 | 4.80| 16  | 36  |
| Age of mothers    | Year  | 33.49 | 4.65| 24  | 43  |
| Age of fathers    |       | 38.07 | 4.14| 30  | 46  |

Abbreviations: M, Mean; SD, Standard deviation; Min, Minimum; Max, Maximum.
### Table 2
Sociodemographic characterization of the participants

| Variable                        | Parent | Frequency | Percent |
|---------------------------------|--------|-----------|---------|
| Number of children in the family|        |           |         |
| One child                       | -      | 24        | 53.3    |
| Two children                    | -      | 19        | 42.2    |
| Three children                  | -      | 2         | 4.5     |
| Education                       |        |           |         |
| High School                     | Father | 7         | 15.6    |
|                                 | Mother | 4         | 8.9     |
| Undergraduate                   | Father | 20        | 44.4    |
|                                 | Mother | 29        | 64.4    |
| Master                          | Father | 11        | 24.4    |
|                                 | Mother | 9         | 20      |
| Doctoral                        | Father | 7         | 15.6    |
|                                 | Mother | 3         | 6.7     |
| Economically active             | Do not Work | 0 | 0     |
|                                 | Mother | 37        | 82.2    |
| Work                            | Father | 45        | 100     |
|                                 | Mother | 8         | 17.8    |

### Table 3
Descriptive statistics of the study measures (n = 45)

| Variable                        | M      | SD    |
|---------------------------------|--------|-------|
| M-CHAT                          | 32.78  | 4.33  |
| GARS-2                          |        |       |
| Stereotypical behaviors         | 13.11  | 7.06  |
| Social interactions             | 16.61  | 8.91  |
| Executive functions: BRIEF      |        |       |
| Inhibition                      | 5.71   | 5.77  |
| Shifting                        | 2.44   | 3.29  |
| Emotional control               | 3.13   | 3.26  |
| Working memory                  | 4.22   | 4.23  |
| Planning /Organization          | 2.29   | 3.11  |
| Self-Control Index (ISCI)       | 8.84   | 8.84  |
| Flexibility Index (FI)          | 5.58   | 5.62  |
| Emergent Metacognition Index (EMI)| 6.51  | 6.84  |
| Global Executive Composite (GEC)| 17.67  | 15.56 |
| Repetitive behaviors            |        |       |
| Stereotyped behaviors           | 10.76  | 3.13  |
| Self-injurious behaviors        | 9.49   | 2.04  |
| Ritualistic behaviors           | 10.69  | 2.63  |
| Sameness behaviors              | 7.82   | 2.02  |
| Restricted behaviors            | 16     | 3.89  |
| Compulsive behaviors            | 7.58   | 3.45  |
| Total score                     | 62.33  | 13.57 |

Abbreviations: M, Mean; SD, Standard deviation
These findings are consistent with the executive dysfunction model that believed RRBs result from executive dysfunction (Hughes et al., 2019). BRIEF-P total score was positively correlated with stereotyped behaviors, self-injurious behaviors, ritualistic behaviors, sameness behaviors, restricted behaviors, compulsive behaviors, and repetitive behaviors total score. The RBS-R total score had the highest correlation with cognitive flexibility and the lowest correlation with planning in executive functions. Neither M-CHAT score nor social interactions score in GARS-2 were correlated with BRIEF-P total score.

### Table 4
Correlation coefficients between variables

| Variables   | Age | ASD | Executive functions | Repetitive behaviors |
|-------------|-----|-----|---------------------|----------------------|
|             |     | M-C | SB | SI | Inhi | Shift | Emoti | Work | Plan | ISCI | FI | EMI | GEC | SB | SIB | RB |
| Age         | 1   | -.39* | -.13 | -.19 | .36* | .14 | .23 | .18 | .20 | .33* | .21 | .20 | .29* | -.12 | -.04 | .24 |
| ASD         | M-C | -.39* | 1   | .53* | .66* | .15 | .16 | 0   | .24 | .11 | .10 | .09 | .19 | .17 | .38** | .17 | .18 |
|            | SB  | -.13 | .53* | 1   | .67** | .36* | .29 | .41** | .36* | .34* | .40** | .41** | .37* | .44** | .66* | .69** | .53* |
|            | SI  | -.19 | .66* | .67** | 1   | .19 | .32* | .18 | .14 | .16 | .20 | .29 | .16 | .24 | .53** | .44** | .51* |
| Executive functions | Inhi | .36* | .15 | .36* | .19 | 1   | .22 | .74** | .73** | .75** | .97** | .56** | .79** | .91** | .50** | .38** | .50* |
|            | Shift | .14 | .16 | .29 | .32* | .22 | 1   | .47** | .15 | .11 | .33* | .86** | .15 | .45** | .16 | .39** | .29 |
|            | Emoti | .23 | 0   | .41** | .18 | .74** | .47** | 1   | .61** | .72** | .89** | .85** | .70** | .87** | .49** | .62** | .48* |
|            | Work | .18 | .24 | .36* | .14 | .73** | .15 | .61** | 1   | .73** | .73** | .44** | .95** | .84** | .53** | .20 | .40* |
|            | Plan | .20 | .11 | .34* | .16 | .75** | .11 | .72** | .73** | 1   | .78** | .49** | .91** | .84** | .40** | .37** | .40* |
|            | ISCI | .33* | .10 | .40** | .20 | .97** | .33* | .90** | .73** | .78** | 1   | .71** | .81** | .96** | .53** | .50** | .52* |
|            | FI   | .21 | .09 | .41** | .29 | .56** | .86** | .85** | .44** | .49** | .71** | 1   | .50** | .77** | .38** | .59** | .45* |
|            | EMI  | .20 | .19 | .37* | .16 | .79** | .15 | .70** | .95** | .91** | .81** | .50** | 1   | .90** | .51** | .29** | .43* |
|            | GEC  | .29* | .17 | .44** | .24 | .91** | .45** | .87** | .84** | .96** | .77** | .90** | 1   | .54** | .47** | .52* |
| Repetitive behaviors | SB  | -.12 | .38** | .66* | .53** | .50** | .16 | .49** | .53** | .40** | .53** | .38** | .51** | .54** | 1   | .57** | .66* |
|            | SIB  | -.04 | .17 | .69** | .44** | .38** | .39** | .62** | .20 | .37** | .50** | .59** | .29** | .47** | .57** | 1   | .56* |
|            | RB   | .24 | .18 | .53** | .51** | .50** | .29 | .48** | .40** | .40** | .52** | .45** | .43** | .42** | .66** | .56** | 1   |
|            | SAB  | .15 | .19 | .47** | .36** | .25 | .56** | .51** | .19 | .29 | .37** | .62** | .25 | .42** | .25 | .54** | .49* |
|            | REB  | .20 | .25 | .47** | .53** | .30** | .59** | .39** | .22 | .12 | .35** | .57** | .19 | .39** | .53** | .42** | .69* |
|            | CB   | -.02 | .20 | .58** | .47** | .27 | .48** | .43** | .31** | .17 | .34** | .53** | .27 | .40** | .58** | .56** | .52* |
|            | RBT  | .09 | .30 | .71** | .62** | .46** | .53** | .60** | .40** | .35** | .54** | .66** | .41** | .57** | .78** | .74** | .84* |

**Abbreviations:** M-C, M-CHAT; SB, Stereotypical behaviors; SI, Social interactions; Inhi, Inhibition; Shift, shifting; Emoti, Emotional control; Work, Working memo ISCI, Self-Control Index; FI, Flexibility Index; EMI, Emergent Metacognition Index; GEC, Global Executive Composite; SB, Stereotyped behaviors; SIB, Self-injurious behaviors; SAB, Sameness behaviors; RB, Restricted behaviors; CB, Compulsive behaviors; RBT, repetitive behaviors total score.

**Correlation is significant at the 0.01 level

*Correlation is significant at the 0.05 level

### Linear Regression Modeling

A linear regression modeling was conducted to prediction and further explore these relationships. For each unit increase in the BRIEF-P total scores, the RBS-R total score increased by 0.33 [t (43) = 4.58, p < .0001]. Likewise, for each unit increase in the BRIEF-P total scores, the stereotyped behaviors score increased by 0.29 [t (43) = 4.17, p < .0001], self-injurious behaviors score increased by 0.22 [t (43) = 3.47, p < .0001], ritualistic behaviors score increased by 0.27 [t (43) = 4.03, p < .0001], sameness behaviors score increased by 0.17 [t (43) = 3.01, p < .004], restricted behaviors score increased by 0.15 [t (43) = 2.77, p < .008], and compulsive behaviors score increased by 0.16 [t (43) = 2.84, p < .008]. There is no significant association between BRIEF-P total score and M-CHAT scores [t (43) = 1.16, p = .25].

### Discussion

The main aim of this study was to examine how EFs in people with ASD are related to the RRBs. We found that increased EFs deficits were related to increased stereotyped behaviors, self-injurious behaviors, ritualistic behaviors, sameness behaviors, restricted behaviors, compulsive behaviors, and repetitive behaviors total score. There were no significant associations between the BRIEF-P total score and M-CHAT score nor social interactions score in GARS-2. These findings are consistent with the executive dysfunction model that believed RRBs result from executive dysfunction (Hughes et al., 1994; Joseph, 1999).
The executive dysfunction models argue that deficits EFs lead to a person being incapable of plan and control behavior, including the ability to inhibit an ongoing response, or spontaneously create a new one. When executive function emerges and develops in toddlers with ASD it is usually expected marked by abnormal cognitive flexibility, response inhibition, and perseveration. Inconsistent with our findings, Lopez et al. (2005) and Mosconi et al. (2009) reported that a specific deficit in the capacity to move from preferred behaviors to new, more adaptive ones contributes to the occurrence of RRBs.

In contrast, there were no relationships between EFs and both M-CHAT score and social interaction ability assessed on the GARS-2. Previous studies also demonstrated and hypothesized that deficits in theory of mind and week central coherency are underlying problems in social functions, whilst impairments in executive functions are underlie RRBs in individuals with ASD (Damasio & Maurer, 1978; Frith & Happe, 2004; Hughes et al., 1994; Joseph, 1999; Turner, 1997).

We also found that parent reports of executive functions predicted the severity of toddlers’ RRBs. This finding is consistent with prior studies (Lopez et al., 2005, Mosconi et al., 2009) that showed EFs appear more closely related to the RRBs. Impairments in EFs have been reported in people with acquired damage to the prefrontal cortex as well as in a range of neurodevelopmental disorders (e.g., ASD) with congenital impairments in this area. Dysfunction of executive functions can explain some core symptoms of ASD, such as rigidity, insistence on sameness, inhibition, and perseveration (Delli et al., 2017). Whereas, other cognitive theories, theory of mind that is the ability of the people to an understanding the social world and apply appropriate behavior or strategy in a social setting are related to social interaction deficits in people with ASD (Baron-Cohen et al., 1985; Delli et al., 2017). According to Alexander Luria's neurobiological processes model, frontal areas are considered as a regulatory brain part and abnormality in this lobe is associated with the disability to follow a sequence of instructions, disinhibition, and repetitive motor movements (Luria, 1969; Luria, 2012). It proposed that impaired EFs contributes to a range of ASD cor symptoms including those observed in the RRBs.

Limitation
Although this research adds information about the relationship between EFs and RRBs, it is not without limitations. First, the cross-sectional study with toddlers under 36 months old limits the opportunity to understand the developmental trajectories of EFs and RRBs in ASD. Second, our EFs and RRBs measures were parent's report instruments. Although the previous study emphasized that parent report is an appropriate research method in the field of the individual with special needs (Fisher et al., 2014), behavioral observations in real-world settings and laboratory tasks can use to enhance the accuracy and validity of data.

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Authors contributions
Data collection, writing the original draft: S.S; study design: S.S, R, Sh, B. HR, P: Writing the original drafts, Approving the final version of the manuscript: All authors.

Conflict of interest
The authors declared no conflict of interest.

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