## ORIGINAL PAPER

# Age Related Differences of Executive Functioning Problems in Everyday Life of Children and Adolescents in the Autism Spectrum

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**Abstract** Numerous studies investigated executive functioning (EF) problems in people with autism spectrum disorders (ASD) using laboratory EF tasks. As laboratory task performances often differ from real life observations, the current study focused on EF in everyday life of 118 children and adolescents with ASD (6–18 years). We investigated age-related and individual differences in EF problems as reported by parents on the Behavioral Rating Inventory Executive Functions (BRIEF: Gioia et al. in Behavior rating inventory of executive function. Psychological Assessment Resources, Odesse 2000), and examined the association with autism severity. Inhibition problems were mostly found in the youngest group (6- to 8-year-olds), whereas problems with planning where more evident for 12- to 14-year-olds as compared to 9- to 11-year-olds. In a subsample of

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A. M. Scheeren · S. Begeer · H. M. Koot Department of Developmental Psychology, VU University, Amsterdam, The Netherlands participants meeting the ADOS ASD cut-off criteria the age related differences in planning were absent, while problems with cognitive flexibility were less apparent in 15- to 18-year-olds, compared to 9- to 11-, and 12- to 14-year olds. EF problems surpassing the clinical cutoff were only observed in 20 % (planning) to 51 % (cognitive flexibility) of the children and adolescents, and no relation was found with ASD symptom severity. This underlines the heterogeneous nature of ASD.

**Keywords** ASD · Autism severity · Behavioral Rating Inventory Executive Functions (BRIEF) · Development · Executive functioning

#### Introduction

The theory of executive dysfunction (Damasio and Maurer 1978; Pennington and Ozonoff 1996) suggests that some autism symptoms might stem from executive functioning

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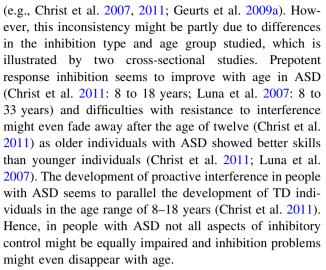
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(EF) deficits. EF refers to cognitive skills that serve independent, purposive, goal-directed, and self-serving behavior (Lezak et al. 2012). A plethora of studies has shown that children, adolescents, and adults with autism spectrum disorders (ASD) encounter problems in executive functioning (e.g., Bramham et al. 2009; Corbett et al. 2009; Sinzig et al. 2008, for reviews see Hill 2004; Russo et al. 2007). Also, EF deficits relate positively to certain autism symptoms (e.g., repetitive behavior: de Vries and Geurts 2012; Yerys et al. 2009). However, children and adolescents with ASD do evolve in their EF skills (Christ et al. 2011; Happé et al. 2006; Luna et al. 2007; Pellicano 2010), and not all individuals with ASD have clinically significant EF deficits (Hill and Bird 2006; Pellicano et al. 2006; Towgood et al. 2009). Hence, within the population of individuals with ASD the development of EF is highly heterogeneous. Moreover, it is important to examine the development of different domains of EF separately, because research in typical development has shown that the structure of EF becomes more differentiated with age (see Hughes et al. 2009; Huizinga et al. 2006; Miyake et al. 2000), and different types of EF develop at a different pace (Best et al. 2009; De Luca et al. 2003; Hughes 2011). Four domains of EF that are traditionally referred to are inhibition, working memory, cognitive flexibility (i.e., shifting), and planning (Hill 2004; Hughes 2011; Pennington and Ozonoff 1996). These four domains have been studied extensively in psychological laboratories in both typically developing (TD) children and children with ASD of varying ages. In the current study we aim to describe developmental EF profiles of children and adolescents with ASD, based on everyday life observations by parents.

The first component of EF, inhibition, refers to the ability to voluntarily and deliberately suppress responses. There are three different types of inhibition (Friedman and Miyake 2001). The first type, inhibition of prepotent responses, refers to the suppression of a dominant response: for example when a child inhibits the response to speak before it is his/her turn. Resistance to interference, the second type, refers to ignoring irrelevant information. This is, for example, required when a child tries to listen to a teacher but hears other children speak. The last type, resistance to proactive interference, refers to processes where previously learned information becomes irrelevant and interferes with new information. This happens, for example, when a teacher tries to learn new names of students, but old names interfere. Improvements of inhibition are evident from childhood to adulthood in typical development (Davidson et al. 2006; Huizinga et al. 2006; Luna et al. 2004). In ASD, deficits have been observed in each of the three inhibition domains in some (e.g., Adams and Jarrold 2011; Christ et al. 2011; Mosconi et al. 2009; Sinzig et al. 2008; Verté et al. 2005), but not all studies



In ASD the developmental trajectory of working memory, the second EF component, seems to differ from the developmental trajectory of inhibition (Luna et al. 2007). Working memory is the ability to maintain and manipulate on-line information (Baddeley 1992). A distinction is made between visual and verbal working memory processes (Smith et al. 1996). In typical development, both components show a linear development between the age of 4 and 15 years (Gathercole et al. 2004), although improvements of visual (spatial) working memory processes are seen even into young adulthood (Luna et al. 2004). Working memory deficits are commonly observed in ASD (e.g., Minshew and Goldstein 2001; Steele et al. 2007, but see Edgin and Pennington 2005), especially in spatial working memory (Williams et al. 2005). Compared to TD individuals, working memory developments seems to be intact in children, but not in adolescents and adults with ASD (Luna et al. 2007). Thus, despite parallel development during childhood, working memory deficits are evident across the lifespan in ASD.

Clear improvements with increasing age are noted in typically developing children and adolescents for the two other EF components, cognitive flexibility (i.e., the ability to intentionally shift thoughts and actions in response to contextual changes: Monsell 2003) and planning (i.e., thinking ahead: Anderson et al. 2001; Davidson et al. 2006; De Luca et al. 2003; Luciana et al. 2009). Problems in both cognitive flexibility and planning have been observed in children with ASD (Hill 2004), but especially for cognitive flexibility the findings are rather inconsistent (Geurts et al. 2009b). However, cognitive flexibility does seem to improve in ASD during childhood (Happé et al. 2006) and development in planning is evident in young children (Pellicano 2010) as well as in young adolescents (Happé et al. 2006) with ASD. In sum, little is known about the developmental patterns of cognitive flexibility and planning in people with ASD, though improvements with age have been observed for these EF domains.



In general, there are three different hypotheses regarding the development of EF in ASD: (1) the development of EF in children and adolescents with ASD might be delayed, but parallel to the typical EF development (Christ et al. 2011; (2) there might be a deviant EF development in ASD (Ozonoff and McEvoy 1994; or (3) a delayed, but parallel EF development in childhood might be followed by a deviant EF development in adulthood (Luna et al. 2007). All of the above might be true given that different EF domains follow other developmental trajectories. This underlines the importance to focus on specific domains instead of the broad construct of EF.

In everyday lives of individuals with ASD behavioral problems are observed that seem related to EF. The Behavioral Rating Inventory Executive Functions (BRIEF: Gioia 2000), a parent questionnaire that is widely used in the clinical practise, addresses those everyday behaviors. Whereas associations are evident with other attention and behavioral problems (BRIEF: Gioia et al. 2000, 2002; McAuley et al. 2010), the BRIEF is only minimally related to laboratory tasks (McAuley et al. 2010). This poses a problem for the association of BRIEF reports to pure, actual EF functioning. This has resulted in some authors arguing that the BRIEF does not actually measure EF (McAuley et al. 2010). Others (Kenworthy et al. 2008) question the ecological validity of laboratory tasks as measurements of EF, arguing that EF related problems in everyday life are observed in people with ASD, even when laboratory task performance is intact. Caution should be made stating that the BRIEF measures actual EF. However, problems described by the BRIEF take place in a social context and are relevant in the everyday lives of people with ASD. Therefore, complementary to what we already know from laboratory task studies, it is important to study these behaviors from a developmental perspective and with respect to individual differences.

Several studies have shown that parents of children and adolescents with ASD consistently report EF deficits on the BRIEF (Boyd et al. 2009; Chan et al. 2009; Endedijk et al. 2011; Kalbfleisch and Loughan 2012; Winsler et al. 2007; Yerys et al. 2009; Zandt et al. 2007). Clinically significant BRIEF scores on different domains are reported in 35-70 % of children with ASD (Gioia et al. 2002; Kenworthy et al. 2005). In a study with 54 children with ASD with a mean age of 11 years inhibition problems were observed in 46 % of the children, working memory problems in 57 %, cognitive flexibility problems in 69 %, and planning problems in 70 % of these children (Kenworthy et al. 2005). The high amount of flexibility problems reported in the BRIEF even discriminates children with ASD from other clinical groups (Gioia et al. 2002). Furthermore, a higher score on the BRIEF Behavioral Regulation Index (containing the subscales inhibit, shift, and emotional control) was associated with more symptoms of communication deficits and restricted repetitive behavior (Kenworthy et al. 2009) as assessed by the Autism Diagnostic Interview-Revised (ADI-R: Lord et al. 1994), the Autism Diagnostic Observation Scale- Generic (ADOS-G: Lord et al. 2000), as well as the Repetitive Behavior Scale-Revised (Boyd et al. 2009). This is in line with the observed relations between cognitive flexibility and repetitive behavior in ASD in studies using laboratory tasks (de Vries and Geurts 2012; Yerys et al. 2009). Thus, despite the weak relations between laboratory task performance and observed behaviors, autism symptomatology is related to EF measured with neuropsychological paradigms as well as to reported EF problems in everyday life. Most studies in which the BRIEF is used, are focussed on index scores. However, since the specific EF domains provide more detailed information than a general EF measure, we focus on the four well-described and studied domains in the current study.

The BRIEF has already been used to study developmental trajectories in TD children (Huizinga and Smidts 2011), and recently also in children and adolescents with ASD (Rosenthal et al. 2013). In a large TD sample (431 boys and 416 girls), BRIEF raw subscale scores of four age groups (5- to 8-; 9- to 11-; 12- to 15-; and 16- to 18-yearolds) were compared (Huizinga and Smidts 2011). Working memory and flexibility mainly seemed to develop before the age of 11 years, inhibition appeared to develop until young adulthood, whereas no development seemed evident with regard to the planning subscale. The findings with regard to inhibition fit the conclusions based on laboratory tasks performance. The working memory and cognitive flexibility findings differ from laboratory tasks studies, as development was only evident in childhood, and not during adolescence. The developmental pattern of planning is in contrast with the observed development of planning skills during childhood and adolescence based on EF laboratory tasks performance. In a comparable ASD study (158 boys and 27 girls, Rosenthal et al. 2013), BRIEF standardized (T) scores of four age groups (5- to 7-; 8- to 10-; 11- to 13-; and 14- to 18-year-olds) were compared. Working memory problems were more severe in 14-18 year olds as compared to 6-7 year olds. So, despite the improved performances on working memory tasks during childhood in ASD, parent reports indicate an increase of working memory problems. This is explained by Rosenthal et al. (2013) by the fact that higher real world demands during adolescence create a larger discrepancy with typical development. However, higher demands from the environment might also produce increasing EF problems in typically developing adolescents, which is not found in the Huizinga and Smidts study (2011). One explanation could be that children and adolescents with



ASD are more vulnerable to changes in environmental demands than typically developing peers. Since no agerelated differences were found in other specific EF domains (e.g. inhibition, cognitive flexibility, and planning), it does not seem to be the case that reported everyday EF problems in general increase during adolescence in ASD. As Rosenthal et al. (2013) compared age-related T-scores, thereby providing information about the relative impairment of participants with ASD compared to a typical age norm; it is hard to relate these findings to the findings of Huizinga and Smidts (2011). Trajectories of reported everyday EF problems in ASD, using raw scores, would provide the information necessary for these comparisons. Furthermore, from the sample of Rosenthal et al. (2013) it is not clear how many children and adolescents actually showed clinically significant problems. Given the heterogeneity of EF problems in ASD (Hill and Bird 2006; Pellicano et al. 2006; Towgood et al. 2009), this information seems highly relevant. Although Rosenthal et al. (2013) controlled for symptomatology, they did not investigate the unique contribution of symptom severity to everyday EF. This would be of interest, given the wide range of ASD severity, and following the theory that some ASD symptoms might stem from EF problems (Pennington and Ozonoff 1996).

The first and primary aim of the current study is to study developmental profiles of specific everyday EF domains in children and adolescents with ASD (6-18 years). For this purpose we use the same cross-sectional approach as was taken in TD children and adolescents (Huizinga and Smidts 2011). Hence, we focus on raw scores instead of T-scores. In line with studies addressing EF in ASD (e.g., Christ et al. 2011; Happé et al. 2006), we expect to observe agerelated improvements of the subscales inhibition and shift. We do not expect to find age-related improvements of working memory and planning, based on the larger BRIEF working memory discrepancy with TD during adolescence in ASD (Rosenthal et al. 2013), and the absence of BRIEF planning improvement in TD (Huizinga and Smidts 2011). Our secondary aims are: to explore the degree of clinically relevant EF problems, and to investigate the impact of ASD severity. Given the heterogeneity of ASD (Hill and Bird 2006; Pellicano et al. 2006; Towgood et al. 2009), we expect some, but not all participants to encounter clinically significant everyday EF deficits. The highest proportions of reported deficits are expected on the shift and planning scale (Gioia et al. 2002; Kenworthy et al. 2009). In an attempt to examine individual differences from a developmental perspective as well, the relative proportions of clinical scores are examined for different age groups. In line with Rosenthal et al. (2013), it is expected that the amount of working memory problems will be significantly higher in children than in adolescents. Finally, we expect to find a positive association between ASD symptom severity and EF deficits, especially with regard to inhibition and cognitive flexibility problems (Boyd et al. 2009; Kenworthy et al. 2009).

## Methods

# **Participants**

The participants, 155 children and adolescents with ASD from 6 to 18 years, took part in a larger study (Scheeren et al. 2010, 2012, 2013) for which they were recruited from a specialized school for normally intelligent pupils with ASD. Inclusion criteria for the present study were threefold. First, ASD was diagnosed by a team of clinicians according to the criteria of the DSM-IV-TR (American Psychiatric Association APA 1994, 2000). The clinicians worked independently from the authors and were blind to the outcomes of the current study. The diagnostic process included an examination of psychiatric, neuropsychological, and speech functioning. Second, clinical diagnoses were verified by a raw score above the Dutch threshold for ASD (which is >60 for boys and >51 for girls) on the Social Responsiveness Scale (SRS: Constantino and Gruber 2007; Dutch version: Roeyers et al. 2011). The SRS is a parent questionnaire with 65 items, ranging from 0 (never true) to 3 (almost always true). It provides an index score for autism symptomatology; a higher total score indicates more autistic traits (Constantino and Gruber 2007). Third, to be included participants had to have a receptive verbal IQ score ≥70 on the Dutch version of the Peabody Picture Vocabulary Test-III (PPVT-III: Dunn and Dunn 2005). After the exclusion of participants with a score below the Dutch SRS threshold score (n = 27), or an IQ < 70 (n = 6), the final sample consisted of 118 participants (16 girls, 102 boys), diagnosed with autism (n = 24), Asperger (n = 14) and PDD-NOS (n = 80). The final sample was divided into four different age groups (i.e. 6- to 8 yearolds, 9- to 11 year-olds, 12- to- 14 year-olds and 15- to 18 year olds: see for similar procedure Huizinga and Smidts 2011). Descriptives of the included participants are presented in Table 1. Supplementary Table 1 provides information about the normed performances on the BRIEF scales, other than those of interest in the current study. Although the ADOS score was used as a predictor in this study, it might concern some that over half of the participants did not score above the ASD threshold on the ADOS. However, all participants are clinically diagnosed within the spectrum, and the SRS was used to confirm this. Therefore, it might be the case that the low ADOS scores indicate a sensitivity problem of the ADOS (Bastiaansen et al. 2011; Gotham et al. 2008). Nonetheless, we repeated



Table 1 Descriptive statistics of the participants

			Groups, a	Groups, age in years (n)	u)								
			Total (118)	3)	6-to 8- (14)	14)	9- to 11- (29)	1- (29)	12- to	12- to 14- (42)		15- to 18- (33)	- (33)
Gender (m/v)			102/16		13/1		24/5		38/4			27/6	
Diagnosis (AU7	Diagnosis (AUT/AS/PDD-NOS)	6	24/14/80		4/1/9		8/1/20		6/1/29			6/5/22	
	M (SD)	Range	M (SD)	Range	M (SD)	Range	M (SD)	Range	M (SD)	Range	$F^*$	d	$\eta_{p}^{2}$
Age	13.1 (2. 9)	6–18	8.1 (.8)	6.4-8.9	10.8 (.9)	9–11.8	13.6 (.8)	12.1–14.8	16.5 (1)	15–18.4	386.50	<.001	.91
PPVT-III	105.3 (12.9)	72–132	105.6 (14.1)	84–126	98.3 (11.3)	72–118	109 (11.7)	80–130	105.9 (13.7)	76–132	3.40	.02	80.
SRS total	87.2 (18.1)	61-133	91.2 (15.5)	67-126	91.3 (17.5)	66-128	87.2 (18.1)	61–133	91.2 (15.5)	67-126	2.61	90:	90.
$ADOS^a$	6.3 (4.1)	0 - 19	7.4 (4.5)	2–19	6 (4.4)	0-18	6.3 (4.1)	0–19	7.4 (4.5)	2–19	.67	.57	.02
Inhibition T	63.1 (11.5)	39–94	65 (7)	54–79	63 (9.4)	43–80	63.1 (11.5)	39–94	(2)	54–79	ı	ı	I
WM T	58.5 (7.8)	40-74	61.4 (8.4)	43–74	58.9 (7)	44–71	58.2 (7.3)	41–73	57.1 (8.9)	40–71	ı	ı	ı
Shift T	64.2 (10.6)	29–88	59.6 (10)	45–81	65.5 (11.5)	29–88	65.2 (9.9)	46–83	63.6 (11)	42–84	ı	ı	ı
Plan T <sup>b</sup>	57.7 (8.7)	36–82	55.3 (6.8)	47–64	55 (8.5)	38–72	(6.8) 5.09	40-82	57.6 (8.7)	36–71	I	ı	ı

Autism Diagnostic Observation Scale Generic communication and social reciprocity

AUT autism, AS asperger, BRI Behavior Regulation Index, BRIEF Behavioral Rating Inventory Executive Functions, IQ intelligence quotient, M mean, PDD-NOS pervasive developmental disorder not otherwise specified, Plan plan/organize, PPVT-III Peabody Picture Vocabulary Test-III, SD standard deviation, SRS Social Responsiveness Scale, T 7 score

\* df = 3,114

<sup>a</sup> df = 3,112/n = 116; <sup>b</sup> n = 117



Table 2 Age-related differences on the BRIEF raw subscale scores

Subscale	Groups, age	Groups, age in years (n)							
	Total $(n = 118)^a$ $M$ (SD)	$6-8$ $(n = 14)^{b}$ $M (SD)$	9–11 (n = 29) <i>M</i> (SD)	12–14 (n = 42) <i>M</i> (SD)	15–18 (n = 33) <i>M</i> (SD)	F	p	$\eta_p^2$	Posthoc* (p)
Inhibit	20.5 (4.7)	23.8 (2.4)	21.1 (3.9)	20.5 (4.6)	18.6 (5.3)	4.59	.01	.11	6-8 > 9-11 (.04), 12-14 (.01), 15-18 (.00)
WM	22.1 (3.9)	24 (4.2)	22.3 (3.5)	22.1 (3.3)	21.2 (4.7)	1.67	.18	.04	_
Shift	17.5 (3.4)	17.3 (2.7)	18.2 (2.9)	17.9 (3.5)	16.3 (3.7)	2.11	.10	.05	_
Plan	25.5 (4.5)	24 (3.7)	23.9 (4.6)	26.9 (4.1)	25.7 (4.8)	3.13	.03	.08	9–11 < 12–14 (.04)

BRIEF behavioral rating inventory executive functions, M mean, Plan plan/organize, SD standard deviation, WM working memory

all analyses with the subsample and describe this sample in supplementary Table 2. When results with regard tot the subsample show different patterns compared to findings concerning the total group this will be mentioned.

With the exception of verbal receptive IQ and age, group descriptives did not significantly differ between age groups. In the ADOS subsample receptive IQ didn't differ significantly between age groups.

#### Measurements

### Behavioral Rating Inventory Executive Functions

The Behavior Rating Inventory of Executive functions (BRIEF: Baron 2000; Gioia et al. 2000; Dutch translation Huizinga and Smidts 2012) is a parent questionnaire that includes 86 statements regarding behavior. Items can be rated on a 3-point frequency scale (1 = never; 2 = sometimes; 3 = often). Raw subscale scores can be calculated for eight subscales (inhibit, working memory, shift, emotional control, initiate, plan/organize, organization of materials, and monitoring). Aggregated subscales provide three index scores: (1) the Behavior Regulation Index: consisting of the scales inhibit, shift, and emotional control; (2) the Metacognition Index: consisting of the working memory, initiate, plan/organize, organization of materials, and monitoring scale; and (3) a total score: a composite of all subscales. For each composed score a higher score means more everyday executive problems. T-scores (M = 50, SD = 10) can be calculated to determine whether scores are potentially clinically significant (T > 65), according to gender- and agespecific norms (Huizinga and Smidts 2012). Please note that, in line with the experimental literature concerning EF, we focused on four relevant BRIEF subscales: inhibit (10 items, e.g., "Interrupts others"); working memory (10 items, e.g., "When given three things to do, remembers only the first or last"); shift (8 items, e.g., "Acts upset by a change in

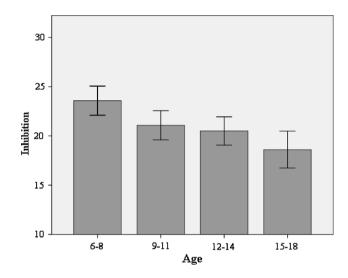


Fig. 1 Mean BRIEF planning subscale scores per age group. The  $\it error\ bars$  represent 95 % CI

plans"); and plan/organize (12 items, e.g., "Does not bring home homework, assignment sheets, materials, and so on"). The BRIEF is a reliable and valid questionnaire (Baron 2000; Gioia et al. 2000, 2002; Huizinga and Smidts 2011).

The Autism Diagnostic Observation Scale Generic

The Autism Diagnostic Observation Scale Generic (ADOS-G: Lord et al. 2000) is a semi-structured instrument used to observe social interaction, communication, imagination and repetitive interests in people with supposed ASD. The developmental and language level determine which one of four modules of approximately 45 min is carried out. Behaviors are scored on a 3-point scale (0 = normal; 1 = slightly abnormal; 2 = clearly abnormal). Scores are calculated for five domains: communication; social reciprocity; fantasy; repetitive interests; and other behaviors. A threshold ( $\geq 7$ ) for the aggregated scores



<sup>\*</sup> Corrected for multiple comparisons

a n for analyses is 117

b n for analyses is 13

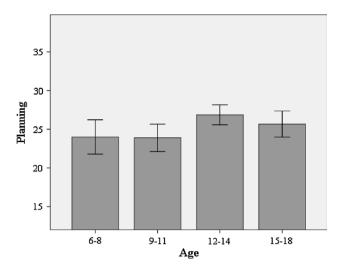


Fig. 2 Mean BRIEF inhibition subscale scores per age group. The  $\it error\ bars$  represent 95 % CI

of communication and social reciprocity determines whether the cut-off for ASD is met. There is a standardized continuous ADOS score (Gotham et al. 2009), which would be most suitable to measure severity. However, module 4 is not standardized. Therefore, we used the ADOS aggregated scores of communication and social reciprocity as a measure of severity. A higher score reflects more autism symptoms. Note, that in the current study module 3 (N = 39) or 4 (N = 77), for children or adolescents with fluent speech, were used. The validity and reliability of the ADOS are reported as good to excellent (Lord et al. 2000).

## Procedure

For each participant there was parental informed consent. Participants from 12 year and older also gave informed consent for themselves. Both the ADOS and BRIEF were administered as part of a larger research project (Scheeren

et al. 2010, 2012, 2013), in which various other questionnaires and experimental tasks were used.

## Outliers and Missing Datapoints

There were no outliers (>3 SE) in the current sample on any of the included BRIEF scales. Following the BRIEF manual (Huizinga and Smidts 2009), if no more than 2 subscale items were missing, missing items were scored as 1 and subscales were computed. SRS missing items were scored as the mean score of the completed subscale items, only when no more than 2 items per scale were missing. The SRS total score was calculated as the sum of the subscales. More than 2 items were missing for one participant on the BRIEF plan/organize scale. Consequently, this score is missing for this participant. The ADOS was only assessed with 116 participants. Therefore, for two participants the ADOS score is missing.

#### Results

Are there Differences Between the Age Groups on the BRIEF Scores?

A multivariate analysis of variance (MANOVA) was conducted with the four BRIEF scale raw scores as dependent variables, and age group as between subject factor. Alpha level for the MANOVA and the subsequent ANOVAs were set on .05. Given the unequal sample sizes and, for inhibition and working memory, the failure to meet the heteroscedasticity criterion (Field 2009), the Games Howell procedure was used in posthoc procedures. Effect sizes were expressed with partial  $\eta^2$ , representing small, medium, and large effects by values of respectively  $\geq$ .01, .06, and .14 (Cohen 1992). In Table 2 results of the MANOVA, and subsequent ANOVAs, were reported.

A significant main effect of age was found, Wilks  $\Lambda = .654$  F(12,291.32) = 4.23, p = < .001, partial

Table 3 Percentages of clinical scores on the BRIEF subscales

Subscale	Groups, age	Groups, age in years (n)										
	Total $(n = 118)$	6-8 (n = 14)	9–11 (n = 29)	12-14 (n = 42)	15-18 (n = 33)	Fisher's exact test	р	Cramer's V				
Inhibit	50.0	48.3	38.1	36.4	41.5	1.58	.68	.11				
WM	50.0	13.8	19.0	24.2	22.9	6.77	.07	.25				
Shift	28.6	51.7	57.1	51.5	50.8	3.41	.34	.17				
Plan <sup>a</sup>	0.0	10.3	33.3	18.2	19.7	9.21	.02	.29				

ADOS autism diagnostic observation scale generic communication and social reciprocity, BRIEF behavioral rating inventory executive functions, M mean, Plan plan/organize, SD standard deviation, WM working memory



n = 117

Table 4 Regression analyses with BRIEF subscales as dependent variables

RRI	HH	cca	00

Predictors	Inhibit			WM			Shift			Plan		
	$\Delta R^2$	β	p	$\Delta R^2$	β	p	$\Delta R^2$	β	P	$\Delta R^2$	β	p
Step 1	.12		.001	.05		.07	.02		.28	.05		.06
Age		35	<.001		22	.02		17	.08		.16	.09
PPVT-III		.05	.61		.08	.41		.01	.95		.14	.14
Step 2	.00		.999	.01		.25	.02		.11	.00		.59
ADOS		.00	.999		11	.25		15	.11		.05	.59
Total $R^2$	.12		.002	.06		.09	.09		.16	.16		.12

Beta's are standardized beta's from the full model

ADOS autism diagnostic observation scale generic communication and social reciprocity, BRIEF behavioral rating inventory executive functions, PPVT-III Peabody Picture Vocabulary Test-III

 $\eta^2=.13$ , indicating differences between the age groups on both the inhibit scale, and the plan/organize scale, but not on the working memory and the shift scale. The follow up analyses revealed that compared to 6- to 8- year-olds, inhibition problems were less severe in the older groups (see Fig. 1).

However, for planning there was another pattern of results (see Fig. 2) as compared to 9- to 11-year-olds, 12- to 14- year-olds showed more planning problems, p = .04. None of the other post hoc comparisons for planning were significant.

As some aspects of IQ cover EF, controlling for IQ might provide overcorrected or unlikely results (Dennis et al. 2009). However, we repeated the analyses with receptive verbal IQ as a covariate: the same pattern was found, except that age-related differences in planning were only moderately significant, F(3,112) = 2.49, p = .06.

In the ADOS cut-off subsample there was a main age effect as well, Wilks  $\Lambda = .487$  F(12,108.77) = 2.83, p = .002, partial  $\eta^2 = .21$ . However, in this sample this was caused by age differences in inhibition, F(4,43) = 2.92, p = .04, partial  $\eta^2 = .17$  (less problems in 15-18- year-olds, compared to 9- to 11- year olds, p = .04) and shift, F(4,43) = 5.37, p = .003, partial  $\eta^2 = .27$  (less problems in 15-18- year-olds, compared to 9- to 11-, p = .04; and 12- to 14- year olds, p = (< .01). When repeating the analyses of the subsample with receptive verbal IQ as covariate the same pattern was found.

What is the Proportion of Children and Adolescents with Clinical BRIEF Scores, and are Proportions Different Between Age Groups?

The BRIEF scores were first categorized as clinical (T > 65) or within normal limits  $(T \le 65)$ . Based on this, the proportions of actual clinical scores per age group were

calculated. Next, Fisher's exact tests with adjusted alpha levels of .01 (.05/4) were used to test differences in relative proportions across age groups. Effect sizes were expressed with Cramer's V. Effects from  $\pm .1$  represent a small effect, this is medium for  $\pm .3$ , and large for  $\pm .5$  (Field 2009). Clinical scores were observed for inhibition in 42 % of the participants, for working memory in 23 %, for shift in 51 % and for plan/organize in 20 % of participants. For the plan/organize scale clinical scores were absent in the youngest group, but present in 33 % of the children from to 9- to 11-years-old. Nonetheless, no significant age group differences were found with regard to clinical percentages of all BRIEF scale scores (see Table 3). In the ADOS cutoff subsample this was 43 % for inhibition, 20 % for working memory, 47 %, for shift, and 21 % for plan/ organize and, again, there were no age differences.

What is the Unique Contribution of ASD Severity to BRIEF Scores?

Four multiple, hierarchical regression analyses were performed with BRIEF subscale scores as dependent variables. As receptive verbal IQ differed between age groups, we included IQ together with age in the first step, and ADOS aggregated scores in the second step. Alpha level was set at .01 (.05/4). Prior to the regression analyses, correlation analyses were performed. Alpha level was set at .01 (05/4). Correlation analyses revealed no relations between the BRIEF subscales and the ADOS: Inhibit, r(114) = .04, p = .70; working memory, r(114) = .1, p = .33; shift, r(114) = .13, p = .16; plan/organize, r (114) = .02, p = .87. In the ADOS cut-off subsample relations were absent as well: Inhibit, r(49) = .04, p = .81; working memory, r(49) = .03, p = .83; shift, r(49) = .02, p = .92; plan/organize, r(48) = .032,p = .84. Regression analysis showed that 12 % of the variance in the inhibit scale was explained. This was 16 %



in the ADOS cut-off subsample. However, in both cases this was fully attributable to the effect of age. Autism severity did not add uniquely to the variance of the inhibition scale, nor did IQ (see Table 4). With respect to the other BRIEF subscales (working memory, shift and plan/organize), none of the regression models were significant. See the results and statistics in Table 4.

### Discussion

In the current cross-sectional study we focused on agerelated differences in specific domains of everyday EF in children and adolescents with ASD as reported by their parents. Moreover, we explored age differences in proportions of clinically significant EF problems, and the relationship between ASD symptom severity and everyday EF. Age-related differences were found with regard to inhibition and planning. Compared to the 6- to 8-year-olds, inhibition problems were reported less for the older children and adolescents. For planning, an opposite pattern was found. Compared to the 9- to 11-year-olds, in 12- to 14year-olds more planning problems were observed. Hence, planning problems may be especially apparent in young adolescents with ASD during the transition period from primary to secondary education. In a subsample of children and adolescent that scored above the ASD threshold on the ADOS, differences were found between age groups for inhibition and cognitive flexibility. Cognitive flexibility problems were less apparent in the oldest group (15- to 18-year olds), compared to 9- to 11-year-olds and 12- to 14- year-olds. Consistent with former studies, everyday EF deficits were found in children and adolescents with ASD, although to a smaller extent than expected. Only 20 % (planning) to 51 % (cognitive flexibility) of the participants encountered clinical EF problems. In the ADOS cut-off subsample this was approximately the same. This highlights the heterogeneity of ASD, and underlines the importance of focusing on individual differences when studying EF in ASD. Counter to our expectation, no relations between ASD symptom severity and everyday EF were found.

## Developmental Profiles of Everyday EF

As expected, in general, older children and adolescents with ASD showed fewer inhibition problems. However, this was only the case when comparing the older age groups with the youngest group. This might suggest a delayed or protracted development of inhibition, rather than a deviated development, which is in line with Rosenthal et al. (2013). Longitudinal research with both ASD and TD participants is necessary to test this hypothesis.

Age-related differences were not found in working memory and cognitive flexibility. With respect to working memory, this contrasts the observed decrease of everyday working memory problems during childhood in TD (Huizinga and Smidts 2011), which consequently fits the increasing T-scores found by Rosenthal et al. (2013). Hence, it might be the case that working memory problems in ASD are specifically revealed during adolescence. Increased real world expectations (Rosenthal et al. 2013) do not seem to explain this though, because age-related differences of raw scores could not be found. Considering that laboratory task performance revealed a deviant development of working memory during adolescence in ASD (Luna et al. 2007) an explanation at the cognitive level might be more valid. Hence, it seems that working memory development in children and adolescents with ASD differs from that of typical development, which, with age, might lead to an increase of impairments compared to TD. Nonetheless, differences between real life observations and observed laboratory performance make it hard to draw firm conclusions. Further research could focus on the unique contribution of specific cognitive developmental processes on the one hand, and increasing environmental demands on the other hand. With respect to cognitive flexibility, the lack of age-related differences in the current study is against our expectations and inconsistent with previously observed age-related improvements in cognitive flexibility performance in ASD (Happé et al. 2006). However, in a subsample of children and adolescents that scored above the ASD threshold on the ADOS we did note age related differences, as the 15- to 18- year olds had fewer problems than the 9- to 11- and 12- to 14-year-olds. One explanation might be that children and adolescents that score above the ASD threshold on the ADOS (and have more severe ASD than those who score below the threshold) receive more professional help or help from their families with switching. However, this is only speculative. In line with Rosenthal et al. (2013), in both of the current samples an increase of flexibility problems was absent. This might suggest that, if cognitive flexibility problems are characteristic for ASD, there is at least no increase of problems during childhood and adolescence. Longitudinal research with ASD and TD participants could test this hypothesis.

Contrary to findings based on laboratory tasks (Happé et al. 2006; Pellicano 2010), in the total group of children and adolescents with ASD, more planning problems were observed in 12- to 14-year-olds compared to 9- to 11-year-olds. This might well be explained by two interdependent factors. First, this could stem from changing demands of the environment. Naturally, children from 12 years and older are expected to behave more independently with regard to school tasks, homework, and everyday activities



than younger children. In the Netherlands it is uncommon for children younger than twelve to receive homework. Although an increase of planning problems in ASD was not found in their study, this is in line with the arguments of Rosenthal et al. (2013) about increased expectations from the environment. Second, parents of younger children might consequently rate behaviors as "never a problem", when these behaviors are not yet asked for in real life. Thus, this does not mean that planning problems are absent, but that the BRIEF might be insufficient to detect these problems. Indeed, the observed age-related differences disappeared in an exploratory analysis with a smaller planning scale where we excluded the items referring to homework or tasks (5 items). To determine whether or not daily life planning abilities change across the life span in children and adolescents with ASD, it is of importance that a planning scale is developed that is more valid with respect to a broader age range. In the ADOS cut-off subsample an increase of planning problems was absent. Again, although speculative, this might be explained by the fact that the family of children and adolescents with more severe ASD or professionals provide more help compared to those supporting participants with milder symptoms, thereby preventing an increase of planning problems when homework is involved.

Summing up, inhibition problems were most apparent in the 6- to 8- year-olds, whereas in the total group studied more planning problems were observed in 12- to 14- year-olds compared to 9- to 11- year-olds. With regard to working memory and cognitive flexibility no age-related differences were found, except that in the ADOS cut-off subsample cognitive flexibility problems were less apparent in the oldest group (15- to 18- year olds), compared to 9- to 11-, and 12- to 14- year-olds. This clearly underlines the importance of studying EF domains separately.

The Proportion of Children and Adolescents with Clinically Significant EF Problems

Compared to previous studies (Gioia et al. 2002; Kenworthy et al. 2005), we found relatively small proportions of children and adolescents with ASD with clinically significant EF problems (inhibition: 42 %; working memory: 23 %; cognitive flexibility: 51 %; planning: 20 %). This could be influenced by the fact that the samples from the previous studies came from a hospital based neuropsychology service, to which children might have been referred because of behavioral problems. The current sample might be more representative for the prevalence of

<sup>1</sup> For this restricted planning scale we eliminated all questions with

<sup>&</sup>lt;sup>1</sup> For this restricted planning scale we eliminated all questions with the words "homework", "task", or "assignment" in the in Dutch translated descriptions, F(3,112) = 1.19, p = .32.



EF deficits in a more general, ASD population with normal to high intelligence levels.

Consistent with other studies, we found that flexibility belongs to the most affected domains of EF (Gioia et al. 2002; Kenworthy et al. 2005). In fact, everyday problems in inhibition, working memory, and planning were only visible in a minority of the children and adolescents with ASD. No age-related differences in proportions of clinically significant scores were observed. This clearly shows that, although group differences exist between groups with ASD and TD (Boyd et al. 2009; Chan et al. 2009; Endedijk et al. 2011; Kalbfleisch and Loughan 2012; Winsler et al. 2007; Yerys et al. 2009; Zandt et al. 2007), not all individuals with ASD encounter EF problems in one or more EF domains.

# The Contribution of ASD Severity

Autism severity, as observed by an objective informant, did not contribute to any of the four EF domains (inhibition, working memory, cognitive flexibility, and planning). To analyse whether the lack of association was due to the relatively mild severity of autistic problems in the sample, we have repeated the analyses with a subsample of participants that scored above the ASD threshold on the ADOS. In this subsample no relation with severity was found either. This is surprising, since the BRIEF is widely used in the clinical practice with regard to ASD, and the ADOS score is based on a broad spectrum of ASD symptoms. Exploratory analyses<sup>2</sup> revealed moderate to strong relations between the SRS and the aforementioned BRIEF scores, while no relation between the ADOS score and the SRS index score was found. Relations between the parent report (SRS) and the BRIEF can be explained by informant- and content-overlap. A lack of relations between the BRIEF and the ADOS might be explained by construct validity problems of the BRIEF. However, it could also be the case that the BRIEF is a more general measurement of behavioral disruption (McAuley et al. 2010) instead of EF. Items like "interrupts others", "gets out of seat at the wrong times, and "gets out of control more than friends" might refer to more generally disruptive and/or inattentive behaviors than to EF. Rather than ASD symptoms, the BRIEF could thus pick up on other symptoms, like comorbid ADHD. This is a question that should be addressed in future research.

Next to the already mentioned shortcomings, one shortcoming of this study is that the youngest age group was too small to detect differences with a medium or small

<sup>&</sup>lt;sup>2</sup> Relations ADOS severity score: SRS index score, r = .07; inhibition, r = .49; working memory, r = .39; shift, r = .86; and planning, r = .56. Data can be obtained from the first author.

effect. It could, therefore, be argued that we had insufficient power to determine age effects. However, we solved this by using more powerful regression analyses with age as a predictor to confirm findings.

To conclude, the BRIEF was developed to measure everyday behaviors that are often found disturbed in children and adolescents with ASD and that seem related to EF. Based on the current study, we wonder whether the BRIEF is an appropriate instrument to measure everyday EF from a developmental perspective, however. The perceptions of parents on behaviors take place in a context of environmental demands that vary with age. It is, therefore, complex to determine whether observed differences between age groups point to true developmental changes of an underlying deficit or simply reflect responses to changing environmental demands. The planning scale in particular seems more appropriate to measure problems in adolescents than in children. Therefore, adjustments might be needed in order to increase ecological validity for a broad age range. In a clinical assessment it would be best to study EF with laboratory tasks as well as the BRIEF questionnaire. When both EF skills and environmental demands are taken into account this might help determining whether interventions are needed to aim at actual EF, the social environment, or both. Furthermore, profiles of deficits might predict developmental (Berger et al. 2003) or intervention outcomes (van der Oord, et al. 2008), which could benefit the efficiency and effectiveness in the clinical practice. Nonetheless, likewise to EF studies that take place in the laboratory, the current study highlights that some, but not all domains of everyday EF are impaired in some, but not all children and adolescents with ASD. Since the majority of the children and adolescents does not seem to have clinically significant problems, even within a subsample of children and adolescent who scored above the ASD threshold on the ADOS, and because autism severity is not related to everyday EF problems it is important to focus on individual differences in ASD.

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