



Behavioral flexibility in children with autism spectrum disorder and intellectual disability



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ABSTRACT

Children with autism spectrum disorder (ASD) have particular difficulty with behavioral flexibility, but the knowledge base on behavioral flexibility in children with a diagnosis of ASD plus intellectual disability (ID) compared to children with ID only is still scarce. The aim of the present study was to assess behavioral flexibility in 111 children (84 boys) with ASD (87 autistic disorder; 24 PDD-NOS) plus ID (IQ range 10.59–72.67) and compare their scores to those of a control group consisting of 65 children with ID only (42 boys). Their age range was between 2;7 and 9;11 years/months. Behavior flexibility was measured using a Dutch version of the Behavioral Flexibility Rating Scale – Revised (Green et al., 2006; Peters-Scheffer et al., 2008). Results showed that behavioral flexibility in children with ASD plus ID was predicted by autism severity, developmental age, and initiating social interaction. A lack of behavioral flexibility seems to influence emotional and behavioral problems and maternal stress, but not adaptive behavior.

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1. Introduction

Children with autism spectrum disorder (ASD) display impairments in social interaction and communication and show a restricted repertoire of activities and interests (American Psychiatric Association, 2000). Several studies associate these restricted and repetitive behaviors and interests with executive dysfunctioning and most clearly with the domain of cognitive flexibility (e.g., Lopez, Lincoln, Ozonoff, & Lai, 2005; South, Ozonoff, & McMahon, 2007). This is defined as the ability to adapt thoughts or actions in response to situational changes (Geurts, Corbett, & Solomon, 2009). In natural settings, deficits in flexibility are frequently reported in individuals with ASD (e.g., Gioia, Isquith, Kenworthy, & Barton, 2002), but laboratory studies using neuropsychological tests such as Wisconsin Card Sorting Test and the Trail Making Test or other experimental cognitive paradigms have yielded inconsistent findings (e.g., Corbett, Constantine, Hendren, Rocke, & Ozonoff, 2008; Hill & Bird, 2006; Lopez et al., 2005; South et al., 2007). Findings might be confounded by characteristics of the participants (e.g., intellectual functioning, verbal ability, age and co-occurring disorders), and task demands such as the explicitness of the task instruction and the amount of disengagement required to perform the switch (Geurts et al., 2009; Van Eylen et al., 2011). As stated by Geurts et al. (2009), based on face-validity, cognitive flexibility seems related to the insistence

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of sameness and behavioral rigidity (i.e., lack of behavioral flexibility) observed in individuals with ASD, but connecting results of the cognitive flexibility measures to behavioral flexibility in everyday situations has been complex.

Next to executive dysfunctioning, alternative explanations for the lack of behavioral flexibility in individuals with ASD have been proposed, including (a) a homeostatic mechanism to reduce over-arousal, (b) an inability to cope with unpredictability, (c) obsessive-compulsive disorder, (d) a desire for self-stimulation, and (e) a lack of central coherence (Green et al., 2006; Turner, 1999). As these 'theories' are not entirely exclusive, it seems plausible that they complement each other in explaining the onset and the maintenance of behavioral flexibility in individuals with ASD (Turner, 1999). The lack of behavioral flexibility is one of the core features of ASD. However, our knowledge base on the nature and extent of behavioral flexibility in individuals with ASD is scarce, especially about situations in which children with ASD show a lack of behavioral flexibility. Ecologically valid measures are required to resolve the paradox between cognitive and behavioral flexibility (Geurts et al., 2009).

One of the few instruments available for assessing behavioral flexibility is the Behavior Flexibility Rating Scale (BFRS) and the Behavioral Flexibility Rating Scale – Revised (BFRS-R), which were developed by Green and her colleagues (Green et al., 2006, 2007; Pituch et al., 2007) for the purpose of identifying specific situations in which individuals with developmental disabilities show an insistence on sameness. Green et al. (2006) administered the BFRS to 726 individuals with autistic disorder, Asperger syndrome and Down syndrome, and developed its factor structure (Pituch et al., 2007), while Didden et al. (2008) added a control group of individuals with non-specific intellectual disability (ID) and a control group consisting of individuals with Angelman syndrome. Results of these studies showed that individuals with autistic disorder and Asperger syndrome showed significantly more problems in behavioral flexibility than individuals with Down syndrome and Angelman syndrome. When diagnosis was controlled for, no significant relationship between behavioral flexibility and gender or age was found.

However, Green et al. (2006) and Didden et al. (2008) used the same participants with ASD and were not able to ensure the representativeness of the sample due to limitations with the data collection methods. As data in Green et al. (2006) were collected using an internet survey, diagnosis of ASD was established through parental report instead of more reliable standardized measures, such as the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DiLavore, & Risi, 2006) or the Autism Diagnostic Interview (Lord, Rutter, & Le Couteur, 1994). This makes verification of the diagnosis of ASD in the earlier studies impossible. Furthermore, in both studies no data were collected on child variables, such as cognitive functioning, adaptive behavior, and autism severity. Therefore, factors that might predict and/or which might influence behavioral flexibility were not investigated.

In light of these limitations, we aimed to improve the aforementioned studies by confirming the diagnosis of ASD and ID by using reliable and standardized measures and by including several child variables to determine which child factors might predict behavioral flexibility in children with ASD. The aim of the present study was to (a) assess behavioral flexibility in children with ASD (i.e., either autistic disorder or PDD-NOS) plus ID and compare them to children with ID only, (b) explore which factors predict and are influenced by behavioral flexibility in individuals with ASD plus ID, and (c) explore differences in behavioral flexibility scores between individuals with autistic disorder plus ID and those with PDD-NOS plus ID.

Turner (1999) divides repetitive and stereotyped behavior of individuals with ASD into higher level and lower level behavior. She suggests that lower level behavior such as manipulation of objects and stereotyped behavior are more frequent in individuals with lower IQ, while higher level behavior such as repetitive language, circumscribed interests, unusual attachments to objects, and the insistence on sameness are more common in individuals with higher IQ. However, in accordance with some other studies, Gabriels, Cuccaro, Hill, Ivers, and Goldson (2005) found a significantly higher prevalence of sameness behavior in children with ASD with low nonverbal IQ when compared to children with a higher nonverbal IQ. Nevertheless, since typically developing children display an insistence on sameness when they are between two and four years of age (Evans et al., 1997), we assumed that behaviors related to behavioral flexibility such as those measured with the BFRS-R require a certain level of development. Within our sample, which included children with a developmental age between 11 and 41 months ($M = 28.70$; $SD = 7.50$), we therefore expected that children with higher developmental age would experience more problems in behavioral flexibility than children with lower developmental age.

Furthermore, it is likely that children with more severe behavioral inflexibility might also experience more difficulties in learning, especially in natural learning environments. Flexibility appears to be a requirement for extracting relevant information from different stimuli (e.g., verbal vs. non-verbal, auditory vs. visual), persons and contexts. For example, Berger, Aerts, van Spaendonck, Cools, and Teunissen (2003) showed that in a group of 30 high functioning adults with ASD improvements in social competence are related to cognitive shifting ability. Consequently, we hypothesized that children with more severe behavioral inflexibility would have lower adaptive behavior scores.

Next, it was hypothesized that as the severity of autism increased, behavioral flexibility would decrease. As the three subtypes of social interaction and communication (i.e., aloof, passive, and active-but-odd; see Wing & Gould, 1979) may refer to distinct subgroups of children with ASD, they were included to further address heterogeneity (Beglinger & Smith, 2001). Children who were classified as active but odd were expected to have few behavioral flexibility issues, while those classified as aloof were expected to have the highest rates of behavioral inflexibility (Castelloe & Dawson, 1993; Wing & Gould, 1979).

A positive relationship between high behavioral flexibility, early social communication skills (i.e., joint attention, behavioral requests, and social interaction) and advanced language ability was expected. In particular, it was hypothesized that children who are responsive, attentive and have high receptive language skills may have a better understanding of their parents' communication and consequently able to anticipate unexpected changes in their environment. Furthermore, children with better expressive skills may express their needs and ask for clarification in ambiguous situations, which may decrease problems in behavioral flexibility.

Finally, we expected that behavioral inflexibility might be burdensome to both the child and the parent. It was hypothesized that children with higher scores on behavioral inflexibility would display more emotional and behavioral problems. Furthermore, mothers of children with problems regarding behavioral flexibility might be experiencing higher levels of maternal stress than mothers of children who are more flexible.

2. Method

2.1. Participants and setting

Participants were 176 children. One hundred and eleven of them (84 boys) represented the experimental group and were diagnosed with ASD and ID (17.1% profound ID, 31.5% severe ID, 30.6% moderate ID, 18.9% mild ID; 1.8% borderline ID). Of the 111, 87 had received a diagnosis of autistic disorder and 24 were classified as having PDD-NOS. The remaining 65 children represented the control group.

Prior to their inclusion, all children in the experimental group had received a diagnosis of ASD and ID from a clinician who was independent of the study and in accordance with the DSM-IV (American Psychiatric Association, 2000) or ICD-10 criteria (World Health Organization, 1992). For all children, this diagnosis was confirmed by the ADOS (Lord et al., 2006), the Childhood Autism Rating Scale (CARS; Schopler, Reichler, & Rothen Renner, 2007), Mullen Scales of Early Learning (MSEL; Mullen, 1995) and the Vineland Adaptive Behavior Scales (VABS; Sparrow, Balla, & Cicchetti, 1984) as assessed by the first author. The Wing Subgroups Questionnaire (WSQ; Castellote & Dawson, 1993) indicated that 79 children had the aloof subtype, while 20 children had the passive subtype and 12 the active-but-odd subtype. Characteristics of the participants of the experimental group are displayed in Table 1.

The remaining children represented the control group, which was comprised of 65 children (42 boys) with non-specific ID ($n = 44$), physical impairment ($n = 12$), Angelman syndrome ($n = 6$), and Down syndrome ($n = 3$). All were between 3 and 9

Table 1

Characteristics of the participants in the experimental group with ASD and ID and Pearson's correlation coefficients for the Behavioral Flexibility Rating Scale – Revised (BFRS-R) total and subscales, and all other measures for children with ASD plus ID ($N = 111$).

	Autistic disorder plus ID ($n = 87$)		PDD-NOS plus ID ($n = 24$)		Pearson's correlations			
	<i>M</i> (<i>SD</i>)	Range	<i>M</i> (<i>SD</i>)	Range	<i>T</i>	<i>O</i>	<i>E</i>	<i>P</i>
Chronological age (years; months)	5; 7 (17.19)	3; 7–9; 0	5; 11 (19.88)	2; 7–9; 11	.05	.01	.08	.10
Cognitive functioning								
Developmental age in months	28.70 (7.50)	11.25–40.50	21.53 (7.57)	4.75–43.25	.41**	.46**	.26**	.17
IQ	45.77 (15.57)	13.55–71.05	32.85 (14.42)	10.59–72.67	.27**	.32**	.15	.08
Non-verbal IQ	48.71 (15.76)	16.87–76.85	37.92 (15.33)	11.18–75.58	.23*	.28**	.11	.04
Adaptive behavior in months								
Composite	22.96 (6.80)	13–45	18.84 (5.47)	11–43	.34*	.36*	.26**	.13
Communication	30.54 (10.28)	17–53	23.26 (9.28)	12–53	.29**	.31**	.22*	.15
Daily Living Skills	25.21 (8.65)	13–47	21.39 (7.98)	11–45	.39**	.40**	.30**	.16
Socialization	26.96 (8.05)	13–44	20.69 (5.26)	11–41	.24*	.26**	.19*	.14
Autism								
ADOS total	9.08 (156)	7–12	16.71 (2.83)	10–24	-.33**	-.32**	-.28**	-.22*
ADOS communication	3.42 (1.35)	2–7	6.37 (1.40)	2–10	-.20*	-.21*	-.13	-.17
ADOS social interaction	6.08 (1.95)	2–11	10.41 (2.33)	6–16	-.42**	-.38**	-.40**	-.28**
CARS	39.17 (6.92)	26–50	41.25 (5.43)	28–53	.30**	.27**	.27**	.19*
Early Social Communication Scales ^c								
Joint attention: initiating	14.06 (9.54)	0–37	5.77 (7.55)	0–35	.19	.19	.14	.16
Joint attention: responding	149.64 (54.20)	41.66–200	80.49 (64.32)	0–200	.41**	.43**	.30**	.27**
Behavioral requests: initiating	26.53 (8.28)	12–43	22.37 (6.60)	2–37	.14	.19	.06	-.08
Behavioral requests: responding	84.02 (23.69)	25–100	62.45 (33.46)	0–100	.37**	.40**	.24*	.15
Social interaction: initiating	3.06 (1.98)	0–7	2.41 (1.75)	0–8	.38**	.42**	.30**	.17
Social interaction: responding	9.12 (3.57)	4–16	6.35 (3.09)	0–18	.37**	.39**	.27**	.23*
Language								
Receptive language (PPVT)	28.17 (5.24)	21–43	24.17 (3.89)	21–39	.29**	.29**	.21*	.16
Receptive language (RDLS) ^a	26.92 (8.40)	14–41	17.65 (6.40)	1–43	.37**	.39**	.27**	.22**
Expressive language (WO) ^b	26.83 (9.12)	14–43	18.27 (7.34)	14–43	.31**	.32**	.25**	.20*
Emotional and behavioral problems ^b								
Total	68.92 (30.96)	19–137	64.15 (23.62)	7–118	.64**	.61**	.57**	.34**
Internalizing	21.46 (11.10)	7–47	20.99 (8.93)	3–40	.64**	.57**	.58**	.40**
Externalizing	24.83 (11.61)	3–45	23.33 (10.61)	1–58	.48**	.51**	.39**	.20*
Maternal stress ^b					.33**	.35**	.24*	.13*

Note. *T* = behavioral flexibility, total scale; *O* = behavioral flexibility toward objects; *E* = behavioral flexibility toward the environment; *P* = behavioral flexibility toward persons; ADOS = Autism Diagnostic Observation Schedule; CARS = Childhood Autism Rating Scale; PPVT = Peabody Picture Vocabulary Test; RDLS = Reynell Developmental Language Scales; WO = vocabulary test of the Schlichting Test for Language Production.

* $p < .05$.

** $p < .01$.

^a $n = 110$.

^b $n = 108$.

^c $n = 95$.

years of age ($M = 5.02$, $SD = 1.99$). Data on the participants of the control group were collected during a study conducted by Peters-Scheffer et al. (2008), which sought to determine the psychometric properties of the BFRS-R, but only children between 3 and 9 years from the earlier study were included in the current study. Although no formal measures of IQ were administered for the control group, all had ID and attended the same (pre)schools as the children with ASD plus ID in the experimental group. Therefore, the experimental group and the control group were considered to be comparable in terms of their cognitive functioning (i.e., level of ID, IQ, and adaptive functioning).

2.2. Instruments

2.2.1. Behavioral flexibility

The Behavior Flexibility Rating Scale – Revised (BFRS-R; Green et al., 2007) is a scale for assessing behavioral flexibility in individuals with developmental disabilities. The BFRS-R is a revised version of the BFRS. In addition to several wording changes, the revision covered the exclusion of one item and the inclusion of two new items. Using a 3-point Likert-type scale, ranging from zero ('not a problem at all') to two ('the situation causes severe problems'), caregivers rate the severity of challenging behavior that are considered to be triggered by specific and unexpected events and changed routines that could be problematic to the individual. Thus higher scores on the total scale and subscales indicate greater behavioral inflexibility. Factor analysis revealed three factors: (a) flexibility toward objects, (b) flexibility toward the environment, and (c) flexibility toward persons. Internal consistency and intrarater and interrater reliability of the total scale were found to be good to excellent (Peters-Scheffer et al., 2008) and the validity was adequate (Green et al., 2008). For more information regarding the BFRS-R, the reader is referred to Green et al. (2007), Green et al. (2008), Ollington, Green, and Sigafoos (2010), and Peters-Scheffer et al. (2008).

2.2.2. Cognitive functioning

The Mullen Scales of Early Learning (Mullen, 1995) were used to assess the cognitive level of the children with ASD and those with ID. Developmental age was calculated as the average developmental age on the four subscales: fine motor, visual reception, receptive language, and expressive language. Since most children were typically too old and/or too low functioning to determine standardized scores, a ratio IQ was calculated using the following formula: developmental age divided by chronological age and multiplied by 100.

2.2.3. Adaptive behavior

Adaptive behavior levels were assessed using the survey form of the Vineland Adaptive Behavior Scales (Sparrow et al., 1984), a semi-structured interview conducted by a trained interviewer with parents. The VABS consists of a composite score and three subscales: communication, daily living skills, and socialization. Age equivalents in months were used in the analyses.

2.2.4. Autism

The Autism Diagnostic Observation Schedule (Lord et al., 2006) is a semi-structured observation of children in a controlled setting, which is used to evaluate social and communicative functioning in individuals suspected of having an ASD. Depending on the language level of the child, one of four developmental modules of the ADOS is administered. A higher score indicates that a child displays more characteristics of autism. Autism severity was measured using the Childhood Autism Rating Scale (Schopler et al., 2007), a 15-item rating scale completed by an observer on a 4-point scale. Scores are summed to obtain a total score with higher scores indicating greater severity of autism. The Wing Subgroup Questionnaire (Castellote & Dawson, 1993) is a questionnaire with 13 behavioral domains (e.g., communication, social approach, play, imitation, motor behavior, resistance to change) on which parents rate their child's behavior on a scale from 0 (never) to 6 (always) for each domain. A summary score is calculated for each subtype and the highest summary score is considered to indicate the child's subtype.

2.2.5. Early communication and language

The Early Social Communication Scales (ESCS; Mundy et al., 2003) is a videotaped semi-structured observational instrument. The scale measures how the child initiates and responds to tasks involving joint attention, as well how the child responds to behavioral requests and social interaction. Toys and activities are used to elicit social and communicative behavior in an ecologically valid context. Higher scores on the subscales indicate better performance. The first author administered the ESCS, and videotapes were scored by four raters, who were unaware of the exact aim of the study including the other scores of the participants. Interrater reliability was assessed using videotaped data from 28.7% of the children and intraclass correlation coefficients between the paired ratings of the 6 subscales ranged from .66 to .73, suggesting good reliability (Cicchetti, 1994).

Receptive language was measured by the comprehension scales of the Dutch version of the Reynell Developmental Language Scales (RDLS; Van Eldik, Van Der Meulen, Van Der Meulen, Schlichting, & Lutje Spelberg, 1995) and the Peabody Picture Vocabulary Test (PPVT; Dunn & Dunn, 1997). The RDLS consist of 87 items divided into 12 sections in which complexity of tasks increases (e.g., Where is the chair?, Place the doll on the chair). The PPVT measures receptive vocabulary as the child needs to identify the picture named by the experimenter through pointing. Expressive language was measured by the vocabulary test of the Schlichting Test for Language Production. This test measures expressive language as the child needs to name objects and pictures (Schlichting, van Eldik, Lutje Spelberg, Van Der Meulen, & Van Der Meulen, 1995).

2.2.6. Emotional and behavioral problems

The Child Behavior Checklist for ages 1.5–5 (CBCL; Achenbach & Rescorla, 2000) is a 99-item questionnaire to assess behavioral and emotional problems in children from 1.5 to 5 years. For each item, the respondent indicates whether it is not true (0), somewhat or sometimes true (1) or very true or often true (2) now or in the past 2 months. Thus, higher scores on the scales represent more emotional and behavioral problems. The CBCL consists of seven small band scales (i.e., aggressive behavior, anxious/depressed, attention problems, emotionally reactive, sleep problems, somatic complaints, and withdrawn), an internalizing, an externalizing, and a total problem scale.

2.2.7. Parental stress

Parental stress was measured by the Dutch version of the Parental Stress Index – short form (the Nijmeegse opvoedingsstress index – verkort, NOSI-K; De Brock, Vermulst, Gerris, & Abidin, 1992). In general, mothers seem to experience more stress than fathers (Dąbrowska & Pisula, 2010) which can complicate between-subject comparison due to the gender effect of the parent. To avoid the confounding effect parent gender, only mothers – as primary caregivers – were asked to complete the NOSI-K. A higher score represents more maternal stress.

2.3. Procedure

The children were identified by approaching local (pre)schools for children with ID in The Netherlands. Schools distributed letters to the parents of children who met the following intake criteria: (a) chronological age between 2 and 10 years, (b) a documented diagnosis of ID and/or ASD as assessed by a psychiatrist or psychologist using psychometrically reliable and valid measures, and (c) children lived at home so that parents were able to provide information about their child. All parents gave their written consent and did not receive any honorarium for their participation.

Once participants were selected, the first author scheduled in-home interviews with the parents to administer the VABS and the CARS. A week before the parental interview parents completed the BFRS-R, the CBCL, the NOSI-K and the WSQ. The questionnaires were sent out by mail along with an information letter with contact information and instructions on how to complete the questionnaires. During the interview parents returned the completed questionnaires to the first author. If not returned during the interview the first author sent a reminder within four weeks.

In the same month during which the interview was held four assessments at the preschool or school of the child were scheduled to administer the MSEL, the ADOS, the ESCS, and the language tests. Tests were administered by the first author in a separate room at the (pre)school.

3. Results

3.1. Between-group analyses

Based on previous findings on the psychometric properties of the BFRS-R (Peters-Scheffer et al., 2008), a total mean and individual means for the three subscales were calculated for children with autistic disorder plus ID, children with PDD-NOS plus ID and children with ID only. Total and subscale means and mean item scores of the three groups are presented in Table 2.

To test for differences between children with autistic disorder plus ID, children with PDD-NOS plus ID and children with ID a univariate analysis of variance (ANOVA) was conducted with the BFRS-R total scale and a multivariate analysis of variance (MANOVA) was performed with scores on the subscales as dependent variables. There was a significant effect of diagnosis on the subscales ($\Lambda = .82$, $F(6, 340) = 5.77$, $p < .001$), but not on the BFRS-R total scale ($F(2, 172) = 1.17$, $p = .31$).

Separate univariate ANOVA's on the subscales did not reveal a significant effect of diagnosis on either the object sub-scale ($F(2, 172) = 2.54$, $p = .08$) or the environment sub-scale ($F(2, 172) = 1.25$, $p = .29$). However, a significant effect of diagnosis was revealed for behavioral flexibility toward the persons sub-scale ($F(2, 172) = 11.21$, $p < .001$). The Games–Howell post hoc test revealed that behavioral flexibility toward persons was significantly higher in children with autistic disorder plus ID than in children with ID ($p < .001$). No differences in behavioral flexibility toward persons were found between children with PDD-NOS plus ID and children with ID ($p = .79$) and between children with autistic disorder plus ID and PDD-NOS plus ID ($p = .16$).

In sum, multivariate analyses indicated that diagnosis significantly affected behavioral flexibility. However, the precise nature of this relation is yet unclear. Therefore, further analyses were conducted in the group of children with ASD plus ID to explore which variables are associated with behavioral flexibility.

3.2. Variables associated with behavioral flexibility

Besides chronological age, diagnosis and behavioral flexibility scores, no additional data were available for the children with ID. Therefore, only children with ASD plus ID were included in the analysis conducted to determine variables associated with behavioral flexibility. First, correlations were calculated between the total score, the subscales of the BFRS-R and variables related to cognitive functioning, adaptive behavior, ASD, early social communication skills, language, emotional and behavioral problems, and maternal stress. Pearson's correlation coefficients revealed significant associations between

Table 2

Mean scores and standard deviations on the Behavioral Flexibility Rating Scale – Revised (BFRS-R) total, the subscales and the items for the total sample ($N = 176$).

Item	Children with autistic disorder and ID ($n = 87$)		Children with PDD-NOS and ID ($n = 24$)		Children with ID ($n = 65$)	
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>
Chronological age in years	5.33	1.77	4.96	1.71	5.02	1.99
Behavioral flexibility (total)	9.59	6.23	11.65	7.35	9.52	5.46
Behavioral flexibility: objects	6.08	3.95	7.08	3.68	5.20	3.14
Behavioral flexibility: environment	2.00	1.84	2.65	2.52	2.34	1.83
Behavioral flexibility: persons	0.60	0.83	1.13	1.29	1.32	0.99
1. Item misplaced	0.51	0.63	0.74	0.69	0.52	0.59
2. Event postponed	0.69	0.75	0.87	0.82	0.51	0.56
3. Move from current location	0.57	0.68	0.70	0.56	0.48	0.56
4. Item deleted/moved	0.36	0.57	0.43	0.59	0.38	0.52
5. Item unavailable	0.89	0.69	1.13	0.69	0.94	0.58
6. Item broken	0.85	0.86	0.83	0.78	0.66	0.57
7. Change in routine	0.68	0.67	0.83	0.72	0.45	0.52
8. Unexpected interaction	0.28	0.52	0.65	0.78	0.65	0.59
9. Separated from group or family	0.32	0.54	0.48	0.73	0.68	0.56
10. Activity interrupted due to broken item	0.91	0.79	0.91	0.79	0.66	0.59
11. Annoying behavior	0.75	0.80	0.75	0.80	0.77	0.61
12. Item is put in wrong place	0.28	0.56	0.28	0.56	0.26	0.51
13. New item added to the environment	0.22	0.42	0.22	0.42	0.42	0.53
14. Activity interrupted before finishing	0.96	0.71	0.92	0.77	0.72	0.57
15. New activity introduced	0.65	0.71	0.40	0.56	0.51	0.53
16. Other uses processions	1.04	0.71	0.98	0.79	0.92	0.51

Table 3

Mean scores on the Behavioral Flexibility Rating Scale – Revised (BFRS-R) total and the subscales specified by autism severity for those children diagnosed with ASD plus ID ($N = 111$).

Autism severity	Behavioral flexibility: total		Behavioral flexibility: objects		Behavioral flexibility: environment		Behavioral flexibility: persons	
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>
First quartile (30–35)	7.05	5.62	4.67	3.10	1.57	1.94	0.24	0.62
Second quartile (36–40)	7.92	4.84	5.15	3.03	1.42	1.55	0.58	0.70
Third quartile (41–43)	10.96	7.65	6.59	4.59	2.48	2.17	1.04	1.13
Fourth quartile (44–53)	12.05	6.17	7.35	3.87	2.73	2.02	0.81	1.02

behavioral flexibility and cognitive functioning (i.e., developmental age, IQ, non-verbal IQ), adaptive behavior, autism severity, emotional and behavioral problems, parental stress and early social communication, and language (see Table 1).

3.3. Autism severity and subtype

Based on percentile scores on autism severity measured with the CARS, children were divided into four groups. Means and standard deviations are displayed in Table 3. A multivariate analysis of variance was conducted with autism severity as an independent variable and the subscale scores as dependent variables. There was a significant effect of autism severity on behavioral flexibility ($\Lambda = .85$, $F(9, 255.69) = 2.01$, $p = .04$). Separate univariate analyses of variance indicated significant effects of autism severity on the behavioral flexibility total score ($F(3, 107) = 4.12$, $p = .01$), the object sub-scale ($F(3, 107) = 3.09$, $p = .03$), the environment sub-scale ($F(3, 107) = 3.18$, $p = .03$) and the persons sub-scale ($F(3, 107) = 3.30$, $p = .02$). Planned contrasts (repeated) revealed no significant differences between severity groups (all $ps > .05$).

Furthermore, as the social subtypes may refer to distinct subgroups, multivariate analyses were performed with the Wing subtype (i.e., aloof, active-but-odd, and passive) as an independent variable and the sub-scales scores as dependent variables. There were no significant differences in behavioral flexibility between subgroups ($\Lambda = .94$, $F(6, 212) = 1.15$, $p = .34$).

At first glance, the significant effect of autism severity on behavioral flexibility seems to contradict the results in which children with autistic disorder plus ID displayed fewer problems in behavioral flexibility than children with PDD-NOS plus ID. However, although groups were similar on most background variables, an independent t -test revealed that, on average, children with PDD-NOS plus ID ($M = 28.70$; $SD = 7.50$) had a significantly higher developmental age than children with autistic disorder plus ID ($M = 21.53$; $SD = 7.57$, $t(109) = 4.12$, $p < .001$). Therefore, the influence of developmental age (as assessed using the four sub-scales of the MSEL) on behavioral flexibility was determined next.

Table 4

Mean scores on the Behavioral Flexibility Rating Scale – Revised (BFRS-R) total and the subscales for all participants with ASD plus ID specified by developmental age ($N = 110$).

Developmental age	<i>n</i>	Behavioral flexibility: total		Behavioral flexibility: objects		Behavioral flexibility: environment		Behavioral flexibility: persons	
		<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>
9–14 months	22	6.14	5.39	3.50	2.96	1.55	1.97	0.50	0.67
15–20 months	20	7.15	6.52	4.45	3.58	1.45	2.01	0.65	0.99
21–26 months	32	11.84	4.42	7.72	3.67	2.38	1.74	0.63	0.91
27–32 months	22	10.68	5.91	6.73	3.74	2.27	1.45	0.73	1.03
33–38 months	12	13.58	6.44	8.25	2.63	3.08	2.71	1.17	1.27
39–44 months	2	18.50	9.19	10.50	3.54	5.00	4.24	1.50	0.71

3.4. Developmental age

Based on developmental age, children with ASD and ID were divided in six subgroups. Children with a developmental age below 9 months ($n = 1$) and above 44 months ($n = 1$) were excluded from the analysis due to small sample sizes. Descriptives are displayed in Table 4. Results of a multivariate analysis on the sub-scales showed that behavioral flexibility is significantly affected by the developmental age of the child ($\Lambda = .71$, $F(15, 281.98) = 2.50$, $p < .01$).

Univariate ANOVA's on the outcome variables revealed a significant effect of developmental age on behavioral flexibility (total; $F(5, 104) = 5.19$, $p < .001$), the object sub-scale ($F(5, 104) = 6.60$, $p < .001$), and the environment sub-scale ($F(5, 104) = 2.45$, $p < .05$). However, the effect of developmental age on the persons sub-scale was not significant ($F(5, 104) = 1.11$, $p = .36$).

Planned contrasts (repeated) revealed significant differences between children with a development age between 15–20 months and 21–26 months on the total score of behavioral flexibility ($t(5) = 2.78$, $p < .01$) and the object sub-scale ($t(5) = 3.34$, $p < .001$). Other repeated contrasts between developmental age groups were not significant (all $ps > .09$). However, when applying a Bonferroni correction to correct for family-wise error ($p < .003$), only the contrast between the children with a developmental age between 15–20 months and 21–26 months on the object scale remained significant.

3.5. Language and early social communication

Fewer problems in behavioral flexibility were expected in responsive and attentive children who had good language skills. Therefore, a hierarchical regression analysis was performed with behavioral flexibility as the dependent variable. Receptive and expressive language and the sub-scales of the early social communication scales (i.e., initiating and responding to joint attention, initiating and responding to behavioral request and initiating and responding to social interaction) were the independent variables.

In the first and second step, the six subscales of the ESCS were entered in a stepwise manner. Only the subscales responding to joint attention and initiating social interaction contributed significantly to the model. In the third step, receptive and expressive language were entered, but these variables did not contribute significantly to the model ($F_{\text{change}}(2, 87) = 0.576$, $p = .56$). Table 5 displays the results at each step (i.e., the unstandardized regression coefficient [B] and standard error of the unstandardized regression coefficient [$SE B$] and the standardized regression coefficient [β]).

R^2 was significantly different from zero at the end of each step. All three models significantly improve the ability to predict behavioral flexibility, with the first model being the best (model 1 $F(1, 90) = 20.92$, $p < .001$; model 2 $F(2, 89) = 17.52$, $p < .001$; model 3 $F(4, 87) = 8.96$, $p < .001$). After step 3, 29.2% of the variance in behavioral flexibility was accounted for.

Table 5

Multiple regression to predict behavioral flexibility from joint attention, social interaction and receptive/expressive language for children with ASD plus ID ($n = 104$).

	Step 1			Step 2			Step 3		
	<i>B</i>	<i>SE B</i>	β	<i>B</i>	<i>SE B</i>	β	<i>B</i>	<i>SE B</i>	β
Constant	6.18	1.07		3.93	1.20		1.56	3.74	
Responding to joint attention	0.04	0.01	.44**	0.04	0.01	.37**	0.03	0.01	.28*
Initiating social interaction				1.14	0.34	.31**	1.12	0.34	.31**
Receptive language (PPVT)							0.07	0.10	.08
Expressive language							0.07	0.19	.05

Note. $R^2 = .19$ for step 1; $\Delta R^2 = .09$ for step 2; $\Delta R^2 = .01$ for step 3.

* $p < .05$.

** $p < .01$.

Table 6

Multiple regression to predict behavioral flexibility from developmental age, severity of ASD, joint attention and initiating social interaction for children with ASD plus ID ($n = 104$).

	Step 1			Step 2			Step 3		
	<i>B</i>	<i>SE B</i>	β	<i>B</i>	<i>SE B</i>	β	<i>B</i>	<i>SE B</i>	β
Constant	1.41	1.84		−16.23	4.54		−15.24	4.37	
Developmental age	0.38	0.08	.46**	0.42	0.07	.52**	0.24	0.10	.29*
Severity of ASD				0.40	0.10	.36**	0.39	0.09	.34**
Responding to joint attention							0.02	0.01	.21
Initiating social interaction							0.84	0.32	.23**

Note. $R^2 = .21$ for step 1; $\Delta R^2 = .13$ for step 2; $\Delta R^2 = .07$ for step 3.

* $p < .05$.

** $p < .01$.

3.6. Overview of variables contributing to behavioral flexibility

To determine the relative contribution of the variables to behavioral flexibility, a hierarchical regression analysis with behavioral flexibility as the dependent variable and developmental age, severity of autism, initiation of social interaction and responding to joint attention as independent variables was performed. Table 6 displays the results at each step.

R^2 was significantly different from zero at the end of each step. All three models significantly improved the ability to predict behavioral flexibility (model 1 $F(1, 93) = 25.00$, $p < .001$; model 2, $F(2, 92) = 23.49$, $p < .001$; model 3, $F(4, 90) = 15.23$, $p < .001$). Although a significant predictor when entered in combination with the other subscales of the ESCS, in combination with developmental age and severity of autism, responding to joint attention no longer contributed significantly to the model. After step 3, in which developmental age, severity of autism, responding to joint attention and initiating social interaction are included, 40.4% of the variance in behavioral flexibility was accounted for.

3.7. Adaptive behavior

As we expected that a lack of behavioral flexibility would have a negative effect on the development of children with ASD, a regression analysis between total behavioral flexibility score as an independent variable and adaptive behavior as a dependent variable was conducted. As adaptive behavior is associated with developmental age (Schatz & Hamdan-Allen, 1995), the average developmental age on the MSEL was entered in the first step. In the second step, the total score on the BFRS-R and the subscales were entered in a stepwise manner. However, as only developmental age contributed significantly to the model, total behavioral flexibility ($t = 0.08$; $p = .93$) and behavioral flexibility toward objects ($t = -0.29$; $p = .77$), toward the environment ($t = 0.98$; $p = .33$), and toward persons ($t = -0.03$; $p = .97$) were excluded from the analysis.

To assess the direct effect of behavioral flexibility on adaptive behavior, a hierarchical regression analysis with adaptive behavior as the independent variable was conducted. In the first step, behavioral flexibility and the subscales were entered stepwise, while developmental age was entered in the second step. Although behavioral flexibility toward objects significantly predicted adaptive behavior in the first step, it no longer contributed significantly to the model when developmental age was entered in the second step. Results of both analyses are displayed in Table 7.

R^2 was significantly different from zero at the end of each step. Both models significantly improve the ability to predict adaptive behavior (model 1: $F(1, 109) = 226.59$, $p < .001$; model 2: $F(2, 108) = 112.39$, $p < .001$). That is, both models explained 68% of the variance. Hence, the associations between behavioral flexibility and adaptive behavior can be explained in terms of developmental age. There is a positive linear relation between developmental age and behavioral inflexibility and between developmental age and adaptive behavior, therefore, when developmental age is not controlled for, significant correlations were found between behavioral flexibility and adaptive behavior (see Table 1). However, when developmental

Table 7

Multiple regression to predict adaptive behavior from developmental age and behavioral flexibility for children with ASD plus ID ($N = 111$).

	Model 1				Model 2		
	<i>B</i>	<i>SE B</i>	β		<i>B</i>	<i>SE B</i>	β
Step 1:				Step 1:			
Constant	5.65	0.99		Constant	16.30	1.01	
Developmental age	0.61	0.04	.82*	Behavioral flexibility: objects	0.56	0.14	.36*
				Step 2:			
				Constant	5.68	1.00	
				Behavioral flexibility: objects	−0.03	0.10	−.02
				Developmental age	0.62	0.05	−.83*

Note. For model 1: $R^2 = .68$. For model 2: $R^2 = .13$ for step 1; $\Delta R^2 = .55$ for step 2.

* $p < .01$.

age is controlled for, no significant correlation was found between behavioral flexibility and adaptive behavior ($r = .01$; $p = .93$).

3.8. Emotional and behavioral problems

To assess the effect of behavioral flexibility on emotional and behavioral problems, a stepwise regression was conducted with behavioral flexibility and the subscales as independent variables and the total score of the CBCL as a dependent variable. The total behavioral flexibility score contributed significantly to behavioral and emotional problems ($B = 2.50$; $SE B = 0.29$; $\beta = .64$; $p < .001$). This model significantly improved the ability to predict behavioral and emotional problems ($F(1, 106) = 74.90$, $p < .001$) with 41.4% of the variance in behavioral and emotional problems accounted for. As the subscales made no contribution to the model, they were excluded from the analysis (behavioral flexibility toward objects: $t = 0.01$, $p = .99$; behavioral flexibility toward environment: $t = 0.40$, $p = .69$; behavioral flexibility toward persons: $t = -0.68$, $p = .50$).

To determine which sub-scales of the CBCL were associated with behavioral flexibility, Pearson's correlations were calculated between the total scale of behavioral flexibility and the subscales of the CBCL. There were significant associations, with small effects between behavioral flexibility and sleep problems ($r = .22$, $p < .05$), attention problems ($r = .27$, $p < .01$) and withdrawn behavior ($r = .19$, $p < .05$), moderate effects between behavioral flexibility and externalizing behavior ($r = .48$, $p < .001$), anxiety ($r = .59$; $p < .001$), somatic complaints ($r = .42$, $p < .001$), and aggression ($r = .58$, $p < .001$). There were large effects between behavioral flexibility and CBCL total ($r = .64$, $p < .001$), internalizing ($r = .64$; $p < .001$) and the emotional reactive subscale ($r = .71$; $p < .001$).

3.9. Maternal stress

To assess the effect of behavioral flexibility on maternal stress, a stepwise multiple regression analysis was performed between the three sub-scales of the BFRS-R as independent variables and maternal stress as the dependent variable to determine which sub-scales contributed significantly to maternal stress.

Only behavioral flexibility toward objects contributed significantly to maternal stress, ($B = 2.47$; $SE B = 0.65$; $\beta = .35$; $p < .001$) with 12.2% of the variance in maternal stress accounted for. This model significantly improved the ability to predict maternal stress, ($F(1, 102) = 14.21$, $p < .001$). As the behavioral flexibility toward the environment ($t = 0.11$, $p = .92$), and behavioral flexibility toward persons ($t = -0.36$, $p = .80$) did not contribute significantly to the model they were excluded from the analysis.

4. Discussion

This study was the first to explore which variables are associated with behavioral flexibility in children with ASD plus ID. In particular, behavioral flexibility scores were assessed in children with autistic disorder plus ID, PDD-NOS plus ID and ID only, using the Dutch version of the BFRS-R (Green et al., 2006, 2007; Peters-Scheffer et al., 2008). Our main finding was that in children with ASD and ID an increase in developmental age and autism severity is associated with more problems in behavioral flexibility. The present study also displays a significant effect of diagnosis on behavioral flexibility. However, the precise nature of this relationship remains unclear as differences between groups might be explained by differences in developmental age between groups. The significant effect of developmental age on behavioral flexibility is in accordance with results found by Bartak and Rutter (1976), in which higher functioning children with ASD had significant more rituals and experienced more problems in adapting to new situations than did lower functioning children. However, the same study reported that lower functioning children displayed more resistance to environmental change than higher functioning children.

The effect of autism severity is in line with studies of Didden et al. (2008) and Green et al. (2006) who found more problems in behavioral flexibility reported in individuals with autistic disorder and Asperger Syndrome than in individuals with Angelman syndrome and Down syndrome. However, Didden et al. (2008) found no significant differences between individuals with non-specific ID and ASD on the total scale of the BFRS-R. The difference between the studies may be attributed to a lack of differentiation in the Didden et al. (2008) study between autistic disorder and PDD-NOS. They also included children, adolescents and adults in their sample. Furthermore, precise data on IQ and level of social functioning in the ID samples are lacking and direct comparison is therefore difficult. However, differences in chronological age and living setting as well as variation in other domains could explain differences in results between both studies.

Although results suggest that behavioral flexibility was predicted by severity of autism, no effect of social subtype (i.e., aloof, passive, and active-but-odd) on behavioral flexibility was found. However, since children with the aloof subtype had a significantly lower developmental age than children with the passive or active-but-odd subtype, the absence of a difference in behavioral flexibility between subtypes might be explained by differences in developmental age.

To assess how (a lack of) behavioral flexibility influences child and family functioning, data were collected on adaptive behavior, emotional and behavioral problems and maternal stress. Although behavioral flexibility did not predict adaptive functioning, results might indicate that a lack of behavioral flexibility predicts emotional and behavioral problems and increased levels of maternal stress. Evidence for an association between variables, however cannot be considered as evidence of causation. Therefore research addressing the question of whether changes in the causal variable (i.e., behavioral

flexibility) precedes changes in the outcome (i.e., maternal stress and emotional and behavioral problems) is warranted. Furthermore, as behavioral problems are a strong predictor of maternal stress (see Peters-Scheffer, Didden, & Korzilius, 2012), scores on maternal stress can be affected by the presence of behavioral problems, the association between behavioral flexibility and maternal stress is unclear. Future research needs to address these issues using a longitudinal design.

Behavioral problems and maternal stress seem to affect the effectiveness of early behavioral intervention (Osborne, McHugh, Saunders, & Reed, 2008; Symes, Remington, Brown, & Hastings, 2005), currently considered as the treatment of choice for children with ASD (Eldevik et al., 2009; Peters-Scheffer, Didden, Korzilius, & Sturmey, 2011; Rogers & Vismara, 2008). Therefore, treatment that focuses on enhancing behavioral flexibility seems critical (Green et al., 2007; Ollington et al., 2010) and should commence early. Consequently, studies regarding variables associated with behavioral flexibility are warranted and useful in developing interventions which may enable children with ASD to enhance their capacities to understand and manage unpredictable and changing situations.

Some limitations of the study should be mentioned. A shortcoming is the lack of formal IQ measures in the control group, as is the heterogeneity of the control group which consists of children with various diagnoses. Furthermore, we measured behavioral flexibility using only one data source (i.e., parental report on the BFRS-R). Although subjective, parents can report information about several situations and about behaviors which might not be observed in a short assessment. However, in vivo assessment of behavioral flexibility in naturalistic settings and multiple informants (e.g., teachers) completing the BFRS-R would have strengthened the study (see for example: Green et al., 2008; Ollington et al., 2012). An additional limitation was the uneven sample sizes, including the small sample size for the group of children with PDD-NOS plus ID compared to those with autistic disorder plus ID, which may have influenced the findings.

Since demographic data on the children with ID only, were unavailable, we were unable to match the participants on demographic characteristics such as developmental age. Thus, differences between diagnostic groups may be influenced by differences in demographic characteristics. Furthermore, we were not able to assess the relationship between behavioral flexibility and other variables in children with ID only and, therefore, it is not possible to determine whether the relation between behavioral flexibility and developmental age and initiations of social interactions is specific for children with ASD or also representative for other diagnostic groups.

Clearly, further research is necessary to extend these preliminary findings on behavioral flexibility in individuals with ASD to enhance the comprehension of parents and professionals about the functioning and behavior of individuals with ASD. Moreover, knowledge about behavioral flexibility and associated factors may contribute to improvements in (early) intervention for individuals with ASD. For example, recent work using the BFRS-R as part of a play-based assessment has highlighted the need for developing interventions that involve problem solving and tolerance building as opposed to accommodating the child's lack of flexibility. This is particularly important as during daily routines and interactions, change is unavoidable (Green et al., 2008; Ollington et al., 2010).

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References

- Achenbach, T. M., & Rescorla, L. A. (2000). *Manual for the ASEBA preschool forms and profiles: An integrated system of multi-informant assessment*. Burlington, VT: University of Vermont Department of Psychiatry.
- American Psychiatric Association. (2000). *Diagnostic and statistical manual of mental disorders: Fourth edition text revision*. Washington, DC: American Psychiatric Press.
- Bartak, L., & Rutter, M. (1976). Differences between mentally retarded and normally intelligent autistic children. *Journal of Autism and Childhood Schizophrenia*, 6, 109–120.
- Beglinger, L. J., & Smith, T. H. (2001). A review of subtyping in autism and proposed dimensional classification model. *Journal of Autism and Developmental Disorders*, 31, 411–421.
- Berger, H. J. C., Aerts, F. H. T. M., van Spaendonck, K. P. M., Cools, A. R., & Teunissen, J. P. (2003). Central coherence and cognitive shifting in relation to social improvement in high-functioning young adults with autism. *Journal of Clinical and Experimental Neuropsychology*, 25, 502–511.
- Castelloe, P., & Dawson, G. (1993). Subclassification of children with autism and pervasive developmental disorder: A questionnaire based on wing's subgrouping scheme. *Journal of Autism and Developmental Disorders*, 23, 229–241.
- Cicchetti, D. V. (1994). Guidelines, criteria, and rules of thumb for evaluating normed and standardized assessment instruments in psychology. *Psychological Assessment*, 6, 284–290.
- Corbett, B. A., Constantine, L. J., Hendren, R., Rocke, D., & Ozonoff, S. (2008). Examining executive functioning in children with autism spectrum disorder, attention deficit hyperactivity disorder and typical development. *Psychiatry Research*, 166, 210–222.
- Dąbrowska, A., & Pisula, E. (2010). Parenting stress and coping styles in mother and fathers of pre-school children with autism and Down syndrome. *Journal of Intellectual Disability Research*, 54, 266–280.
- Didden, R., Sigafoos, J., Green, V. A., Korzilius, H., Mouws, C., Lancioni, G. E., et al. (2008). Behavioral flexibility in individuals with Angelman syndrome, Down syndrome, non-specific intellectual disability and autism spectrum disorder. *Journal of Intellectual Disability Research*, 52, 530–539.
- De Brock, A. J. L. L., Vermulst, A. A., Gerris, J. R. M., & Abidin, R. R. (1992). *Nijmeegse ouderlijke stress index*. Lisse: Swets en Zeitlinger.
- Dunn, L., & Dunn, L. (1997). *Peabody Picture Vocabulary Test – Revised*. Circle Pines, MN: American Guidance Services.
- Eldevik, S., Hastings, R. P., Hughes, J. C., Jahr, E., Eikeseth, S., & Cross, S. (2009). Meta-analysis of early intensive behavioral intervention for children with autism. *Journal of Clinical Child and Adolescent Psychology*, 38, 439–450.
- Evans, D. W., Leckman, J. F., Carter, A., Reznick, J. S., Henshaw, D., King, R. A., et al. (1997). Ritual, habit, and perfectionism: The prevalence and development of compulsive-like behavior in normal young children. *Child Development*, 68, 58–68.

- Gabriels, R. L., Cuccaro, M. L., Hill, D. E., Ivers, B. J., & Goldson, E. (2005). Repetitive behaviors in autism: Relationships with associated clinical features. *Research in Developmental Disabilities*, 26, 169–181.
- Geurts, H. M., Corbett, B., & Solomon, M. (2009). The paradox of cognitive flexibility in autism. *Trends in Cognitive Sciences*, 13, 74–82.
- Gioia, G. A., Isquith, P. K., Kenworthy, L., & Barton, R. M. (2002). Profiles of everyday executive function in acquired and developmental disorders. *Child Neurology*, 8, 121–137.
- Green, V. A., Sigafoos, J., Didden, R., O'Reilly, M. F., Lancioni, G. E., Ollington, N., et al. (2008). Play-based assessment of tolerance for frustration and problem-solving skills in children with developmental delays. In A. T. Waddell & R. M. McBride (Eds.), *New research on early childhood education*. Nova Science Publishers: New York.
- Green, V. A., Sigafoos, J., O'Reilly, M., Pituch, K., Didden, R., Lancioni, G., et al. (2007). Behavioral flexibility in individuals with autism: Theory, assessment, and intervention. In L. Zhao (Ed.), *Autism research advances* (pp. 63–77). New York: Nova Science Publishers.
- Green, V. A., Sigafoos, J., Pituch, K. A., Itchon, J., O'Reilly, M., & Lancioni, G. (2006). Assessing behavioral flexibility in individuals with developmental disabilities. *Focus on Autism and Other Developmental Disabilities*, 21, 230–236.
- Hill, E. L., & Bird, C. M. (2006). Executive processes in Asperger syndrome: Patterns of performance in a multiple case series. *Neuropsychologia*, 14, 2822–2835.
- Lopez, B. R., Lincoln, A. J., Ozonoff, S., & Lai, Z. (2005). Examining the relationship between executive functions and restricted, repetitive symptoms of autistic disorder. *Journal of Autism and Developmental Disorders*, 35, 445–460.
- Lord, C., Rutter, M., DiLavore, P. C., & Risi, S. (2006). *Autism Diagnostic Observation Schedule*. Los Angeles: Western Psychological Service.
- Lord, C., Rutter, M., & Le Couteur, A. (1994). Autism Diagnostic Interview-revised: A revised version of a diagnostic interview for caregivers of individuals with possible pervasive developmental disorders. *Journal of Autism and Developmental Disorders*, 24, 659–685.
- Mullen, E. M. (1995). *Mullen scales of early learning: AGS edition*. Minneapolis: Pearson.
- Mundy, P., Delgado, C., Block, J., Venezia, M., Hogan, A., & Seibert, J. (2003). *A manual for the Abridged Early Social Communication Scales (ESCS)* Retrieved from: http://www.ucdmc.ucdavis.edu/mindinstitute/ourteam/faculty_staff/ESCS.pdf.
- Ollington, N., Green, V. A., & Sigafoos, J. (2010). Behavioural flexibility in children with autism. In J. Low & P. Jose (Eds.), *Lifespan development: New Zealand perspectives* (2nd ed.). AK, NZ: Pearson.
- Ollington, N., Sigafoos, J., Green, V. A., O'Reilly, M. F., Lancioni, G. E., & Didden, R. (2012). Functional assessment of insistence on sameness in an 11-year old boy with Asperger syndrome. *Developmental Neuropsychology*, 15, 154–159.
- Osborne, L. A., McHugh, L., Saunders, J., & Reed, P. (2008). Parenting stress reduces the effectiveness of early teaching interventions for autistic spectrum disorders. *Journal of Autism and Developmental Disorders*, 38, 1092–1103.
- Peters-Scheffer, N. C., Didden, R., Green, V. A., Sigafoos, J., Korzilius, H., Pituch, K., et al. (2008). The Behavioral Flexibility Rating Scale – Revised (BFRS-R): Factor analysis, internal consistency, interrater and intrarater reliability and convergent validity. *Research in Developmental Disabilities*, 29, 398–407.
- Peters-Scheffer, N. C., Didden, R., & Korzilius, H. (2012). Characteristics of children with autism spectrum disorders and intellectual disability predicting maternal stress. *Research in Autism Spectrum Disorders*, 6, 696–706.
- Peters-Scheffer, N., Didden, R., Korzilius, H., & Sturmey, P. (2011). A meta-analytic study on the effectiveness of comprehensive ABA-based early intervention programs for children with Autism Spectrum Disorders. *Research in Autism Spectrum Disorders*, 5, 60–69.
- Pituch, K. A., Green, V. A., Sigafoos, J., Itchon, J., O'Reilly, M., Lancioni, G. E., et al. (2007). Factor structure of the behavior flexibility rating scale (BFRS). *Research in Autism Spectrum Disorders*, 1, 55–76.
- Rogers, S. J., & Vismara, L. A. (2008). Evidence-based comprehensive treatments for early autism. *Journal of Clinical Child and Adolescent Psychology*, 37, 8–38.
- Schatz, J., & Hamdan-Allen, G. (1995). Effects of age and IQ on adaptive behavior domains for children with autism. *Journal of Autism and Developmental Disorders*, 25, 51–60.
- Schlichting, J. E. P. T., Van Eldik, M. C. M., Lutje Spelberg, H. C., Van Der Meulen, S. J., & Van Der Meulen, B. F. (1995). *Schlichting test voor taal productie*. Nijmegen: Berkhout.
- Schopler, E., Reichler, R. J., & Rothen Renner, B. (2007). *The Childhood Autism Rating Scale*. Los Angeles: Western Psychology Services.
- South, M., Ozonoff, S., & McMahon, W. M. (2007). The relationship between executive functioning, central coherence, and repetitive behaviors in the high-functioning autism spectrum. *Autism*, 11, 437–451.
- Sparrow, S., Balla, D., & Cicchetti, D. (1984). *Vineland Adaptive Behavior Scales: Interview Edition*. Circle Pines, MN: American Guidance Service.
- Symes, M. D., Remington, B., Brown, T., & Hastings, R. P. (2005). Early intensive behavioral intervention for children with autism: Therapists' perspectives on achieving procedural fidelity. *Research in Developmental Disabilities*, 27, 30–42.
- Turner, M. (1999). Annotation: Repetitive behavior in autism: A review of psychological research. *Journal of Child Psychology and Psychiatry*, 40, 839–849.
- Van Eldik, M. C. M., Schlichting, J. E. P. T., Lutje Spelberg, H. C., Van Der Meulen, B. F., & Van Der Meulen, S. J. (1995). *Reynell Test voor Taalbegrip handleiding*. Nijmegen: Berkhout.
- Van Eylen, L., Boets, B., Steyaert, J., Evers, K., Wagemans, J., & Noens, I. (2011). Cognitive flexibility in autism spectrum disorder: Explaining the inconsistencies? *Research in Autism Spectrum Disorders*, 5, 1390–1401.
- Wing, L., & Gould, J. (1979). Severe impairments of social interaction and associated abnormalities in children: Epidemiology and classification. *Journal of Autism and Developmental Disorders*, 9, 11–29.
- World Health Organization. (1992). *The ICD-10 classification of mental and behavioral disorders: Clinical descriptions and diagnostic guidelines*. Geneva: World Health Organization.