Research in Autism Spectrum Disorders 7 (2013) 699–709

Contents lists available at SciVerse ScienceDirect

Research in Autism Spectrum Disorders

Journal homepage: http://ees.elsevier.com/RASD/default.asp

Behavioral flexibility in children with autism spectrum disorder and intellectual disability

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ARTICLE INFO

Article history:
Received 25 February 2013
Accepted 26 February 2013

Keywords:
Behavioral flexibility
Autism spectrum disorder
Intellectual disability
BFRS-R

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.

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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http://dx.doi.org/10.1016/j.rasd.2013.02.016

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 non-verbal 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.