

Confirmatory factor analysis of the BRIEF2 in a sample of youth with Down syndrome

A. Soltani,¹ E. K. Schworer,^{2,3} ID L. A. Jacobson,⁴ M. M. Channell,⁵ N. R. Lee,⁶ G. G. Faight,⁷ ID R. Grzadzinski,⁸ D. Fidler⁹ ID & A. J. Esbensen^{2,10} ID

¹ Department of Educational Psychology, Kerman Branch, Islamic Azad University, Kerman, Iran

² Division of Developmental and Behavioral Pediatrics, Cincinnati Children's Hospital Medical Center, Cincinnati, OH, USA

³ Waisman Center, University of Wisconsin-Madison, Madison, WI, USA

⁴ Department of Neuropsychology, Kennedy Krieger Institute; Department of Psychiatry and Behavioral Sciences, Johns Hopkins University, Baltimore, MD, USA

⁵ Department of Speech and Hearing Science, University of Illinois at Urbana-Champaign, Champaign, IL, USA

⁶ Department of Psychological and Brain Sciences, Drexel University, Philadelphia, PA, USA

⁷ Department of Psychology, The University of Alabama, Tuscaloosa, AL, USA

⁸ Carolina Institute for Developmental Disabilities, University of North Carolina, Carrboro, NC, USA

⁹ Department of Human Development and Family Studies, Colorado State University, Fort Collins, CO, USA

¹⁰ Department of Pediatrics, University of Cincinnati College of Medicine, Cincinnati, OH, USA

Abstract

Background The factor structure of the Behavior Rating Inventory of Executive Function, second edition (BRIEF2) has been widely examined in both typically developing children and specific clinical samples. Despite the frequent use of the BRIEF2 for measuring executive functioning in individuals with Down syndrome, no study has investigated the factorial validity or dimensionality of the BRIEF2 in this population. This study aimed to address this notable gap in the literature.

Methods Parents of 407 children and youth with Down syndrome aged 6–18 years completed the BRIEF2 as part of different studies led by six sites. Three competing models proposed by previous studies were analysed using Confirmatory Factor Analysis: the theoretical structure of the BRIEF2 where the

scales were constrained to load on three factors labelled as Cognitive, Behavioral, and Emotional Regulation, a two-factor correlated model with the merged Behavioral and Emotional regulation, and a single-factor model.

Results The three-factor model provided a better fit than the one- and two-factor models, yet a large correlation was observed between Behavioural and Emotional regulation factors. The results provide meaningful explanatory value for the theoretical structure of the BRIEF2. However, the Behavioral and Emotional regulation factors might be less differentiated and the two-factor structure of the BRIEF2 may also make theoretical and empirical sense.

Conclusions Although more studies are needed to further examine the factor structure of the BRIEF2 in youth with Down syndrome, this investigation provides preliminary support for the interpretation of the three executive function index scores provided by the BRIEF2: Cognitive, Behavioral, and Emotional Regulation.

Correspondence: Anna J. Esbensen, Cincinnati Children's Hospital Medical Center, Division of Developmental and Behavioral Pediatrics, MLC 4002, 3430 Burnet Avenue, MLC 4002, Cincinnati, OH 45229, USA. (e-mail: anna.esbensen@cchmc.org).

© 2022 The Authors. Journal of Intellectual Disability Research published by MENCAP and International Association of the Scientific Study of Intellectual and Developmental Disabilities and John Wiley & Sons Ltd.

This is an open access article under the terms of the Creative Commons Attribution-NonCommercial-NoDerivs License, which permits use and distribution in any medium, provided the original work is properly cited, the use is non-commercial and no modifications or adaptations are made.

Keywords BRIEF2, CFA, Down syndrome, Executive functioning, Trisomy 21

Introduction

Down syndrome (DS) is the most common genetic cause of intellectual disability, occurring in about 15 per 10,000 live births (Llewellyn *et al.* 2019; Mai *et al.* 2019). Most individuals with DS have an extra chromosome 21, which impacts physical, physiological, cognitive, language, and motor development (Esbensen & MacLean 2017). Difficulty with executive functioning (EF) is a key cognitive feature in children and adults with DS. EF refers to several higher-order cognitive processes responsible for controlling and regulating behaviour. Some well-known EF processes are inhibitory control, shifting between thoughts and actions, storing and processing information simultaneously (working memory), control of emotion, planning, organising, and novel response generation (Diamond 2013).

Several studies have identified challenges with EF among children and adolescents with DS relative to their developmental level (Daunhauer *et al.* 2014; Daunhauer *et al.* 2017; Daunhauer *et al.* 2020; de Weger *et al.* 2021; Iralde *et al.* 2020; Lanfranchi *et al.* 2010; Manrique-Niño *et al.* 2020; Schworer *et al.* 2022; Tungate & Connors 2021). Furthermore, distinct patterns of strengths and weaknesses in EF have been reported among individuals with DS (Daunhauer *et al.* 2014; Lee *et al.* 2011; Loveall *et al.* 2017). Relative strengths are reported for emotional control and inhibitory control, while working memory, planning, and organising are areas of relative weakness (Daunhauer *et al.* 2014; Loveall *et al.* 2017). Shifting is reported to be a relative strength at younger ages and a relative weakness at older ages (Daunhauer *et al.* 2014, Loveall *et al.* 2017). EF challenges in persons with DS are associated with poorer academic achievement (Will *et al.* 2017), adaptive behaviours (Daunhauer *et al.* 2017; Esbensen *et al.* 2021; Sabat *et al.* 2020; Tomaszewski *et al.* 2018; Will *et al.* 2021), communication (Kristensen *et al.* 2022; Maiman *et al.* 2017), and social behaviour (Amadó *et al.* 2016).

The growing interest in the impact of EF on individuals with DS highlights the need to evaluate the psychometric properties of a parent-report

measure of daily EF: the Behavior Rating Inventory of Executive Function, second edition (BRIEF2) (Gioia *et al.* 2015) recommended for and commonly used with individuals with DS (Daunhauer *et al.* 2014; Daunhauer *et al.* 2017; Esbensen *et al.* 2017; Esbensen *et al.* 2019; Esbensen *et al.* 2021; Lee *et al.* 2011; Lee *et al.* 2015; Loveall *et al.* 2017; Manrique-Niño *et al.* 2020; Tomaszewski *et al.* 2018). Some psychometric properties of the BRIEF2 have been evaluated, including internal consistency and inter-rater reliability (Esbensen *et al.* 2019). However, the underlying factor structure has not been evaluated in youth with DS.

Development of the BRIEF and BRIEF2

The original BRIEF was initially developed by Gioia (2000) to measure multiple aspects of EF as manifested in everyday life in children and adolescents from 5 to 18 years old. It consists of parent and teacher forms, both of which contain 86 items rated on a 3-point ordinal scale. Scales include Inhibit, Shift, Emotional Control, Initiate, Working Memory, Plan/Organize, Organisation of Materials, and Monitor (Gioia 2000). Conducting an exploratory factor analysis of the eight scales comprising the BRIEF, Gioia (2000) found a two-factor model that was the most theoretically and statistically sound within parent and teacher forms for typically developing children and a mixed clinical sample. The factors represent two general domains of EF: Behavioral Regulation and Metacognition. The Behavioral Regulation Index (BRI) was defined by Inhibit, Shift, and Emotional Control, and the Metacognition Index encompassed Initiate, Working Memory, Plan/Organize, Organisation of Materials, and Monitor (Gioia 2000). These indices combine to obtain a general overall score, the Global Executive Composite.

Later, an updated version of the BRIEF, the BRIEF2, was developed (Gioia *et al.* 2015). Modifications from the original measure included reducing the number of items from 86 to 63, separating the Monitor scale into the Self-Monitor and Task-Monitor scales, and generation of a three-factor model supported by factor analyses. The Metacognitive Index was renamed to Cognitive Regulation Index (CRI), and the BRI was broken down into two distinct indices, BRI and Emotion

Regulation (ERI) (Dodzik 2017; Hendrickson & McCrimmon 2018).

Factor structure of the BRIEF and BRIEF2 in clinical samples

Using confirmatory factor analysis, Gioia *et al.* (2002) initially examined one-, two-, three-, and four-factor models of the BRIEF with a mixed clinical sample of children with ADHD, autism spectrum disorder (ASD), learning disabilities, Tourette syndrome, mood disorders, and seizure disorders. In the one-factor model, all scales were constrained to load on one latent factor. In the two-factor model, the scales were reflected by two latent factors labelled BRI and Metacognition (or CRI). In the three-factor model, some scales of the BRI were constrained to load on an additional latent factor labelled as ERI. Finally, the four-factor model was similar to the three-factor model except the index tapping cognitive regulation (CRI) was parsed into two separate factors. Results revealed that the best fitting model was the three-factor model (Gioia & Isquith 2002). The factor structure of the BRIEF was further evaluated within specific clinical samples, with various models mostly proposed by Gioia and Isquith (2002). For example, in a sample of children and youth with epilepsy, the two-factor solution provided a better fit than either the one-factor or the three-factor model. Likewise, in a group of children and adolescents with ADHD, the two-factor model, with the Monitor scale loading on both factors, provided an acceptable fit to the data as opposed to the one-factor model (Lyons Usher *et al.* 2016; Slick *et al.* 2006). Similarly, using exploratory factor analysis, the two-factor structure was identified for the BRIEF in a sample of children with intellectual disabilities (Memisevic 2015). In contrast, the three-factor model of the BRIEF as opposed to the one- and two-factor models was supported in a sample of adolescents with mild traumatic brain injury (Lace *et al.* 2019).

Even more recently, the proposed factor structures (one-, two-, and three-factor models) were examined with some mixed and specific clinical samples by using the updated version of the BRIEF – the BRIEF2 (Jacobson *et al.* 2020; Lace *et al.* 2021; Parhoon *et al.* 2022). Using CFA, Lace *et al.* (2021) reported that the bifactor two-factor structure of the BRIEF2 (CRI & BRI/ERI) provided the best fit in a

mixed clinical sample of children with ASD, cerebral palsy, brain tumours, unspecified neurodevelopmental disorders, ADHD, psychiatric (e.g. anxiety and depression) disorders, intellectual disabilities, and specific learning disorders. However, using CFA in a large mixed clinical sample of children with ADHD, adjustment disorders, anxiety disorders, encephalopathy, oncologic diseases, epilepsy, and oppositional defiant disorder or conduct problems, Jacobson *et al.* (2020) reported the three-factor model of the BRIEF2 (CRI, BRI, and ERI) demonstrates the best fit, although analyses including items, scales, and indices suggested not more than six interpretable scales. Likewise, in a sample of children with ADHD, results of a CFA showed that the three-factor structure of the BRIEF2 (CRI, BRI, and ERI) was the best fitting model (Parhoon *et al.* 2022).

In summary, there are inconsistent results regarding the factor structure of the BRIEF and BRIEF2 with mixed clinical samples. While some studies supported the three-factor structure of the BRIEF (Gioia & Isquith 2002; Lace *et al.* 2019) and the BRIEF2 (Jacobson *et al.* 2020; Parhoon *et al.* 2022), others reported that two-factor model best explains the factor structure of the BRIEF (Lyons Usher *et al.* 2016; Memisevic 2015; Slick *et al.* 2006) and BRIEF2 (Lace *et al.* 2021). Such inconsistency may reflect the unique EF structure in specific clinical samples. Indeed, due to specific EF profiles in different developmental disorders, such as DS, EF structures may be expressed differently (Ozonoff & Jensen 1999). Hence, further investigation is required to examine the factor structure of the BRIEF2 in specific populations to support its clinical utility (Gioia 2000; Gioia *et al.* 2015; Gioia & Isquith 2002).

Present study

Given the unique pattern of strengths and challenges of EF in DS, evaluation of the factor structure of the BRIEF2 is warranted in this population to support clinical practice and outcome measurement in research studies. This study aimed to address this notable gap in the literature by examining different competing models of the BRIEF2 structure in youth with DS using CFA. Specifically, the present study examined one-, two-, and three-factor competing models of the underlying structure of the BRIEF2 parent form.

Method

Study design

Data from the current study were combined from six studies led by sites across the United States, and all study procedures were approved by their respective site Institutional Review Boards (IRBs). Data sharing or data use agreements were also obtained. The studies led at the six sites included community based in-person single-site and multi-site studies as well as a national online survey. The age ranges of the studies varied; only data for youth ages 6–18 years were pooled for analyses. Sites spanned the Western, Southern, Mid-West, and Eastern United States, with the online survey being disseminated nationally.

Youth were required to have a diagnosis of DS. The child's demographics and ratings of daily EF abilities were provided by parents. Child cognition was assessed using the KBIT-2 ($n = 180$), KBIT-2 nonverbal ($n = 30$), Leiter-3 ($n = 38$), SB5 ($n = 7$), or DAS-II ($n = 24$). The IQ composite scores for one site were not available, as the study was a national online survey ($n = 121$). In addition, there were no available IQ scores for seven participants in other sites. As IQ was not a key construct in the study design and only used for descriptive purposes, this missingness does not impact interpretation of results.

Participants

This study included 407 youth with DS and their caregivers. Youth with DS were 6–18 years old ($M = 12.16$, $SD = 3.29$) with 126 (31%) children between 6–9 years old, 153 (37.6%) between 10–13 years old, and 128 (31.4%) between 14–18 years old. Sex was approximately equal in the sample with 209 (51.4%) males and 198 (48.6%) females. Most participants reported their race as White (86.7%), with 4.9% Black, 4.2% Asian, 0.3% American Indian, and 3.9% from other races. Ninety per cent were not Hispanic or Latino, 9.8% were Hispanic or Latino, and ethnicity was not reported for one participant (0.2%). Standardised IQ composite scores were available for 279 participants and ranged from 25 to 84 ($M = 51.18$, $SD = 11.55$) with an outlier ($IQ = 90$).

Measures

Executive functioning

Behavior Rating Inventory of Executive Function, second edition (BRIEF2). The BRIEF2 parent form (Gioia *et al.* 2015) is a measure assessing day-to-day EF challenges in children and youth ages 5 to 18 years in their home environment. The measure encompasses 63 items divided among 9 scales: Inhibit, Self-Monitor, Shift, Emotional Control, Initiate, Working Memory, Plan/Organize, Task-Monitor, and Organisation of Materials. On the measure, EF challenges are rated on a 3-point scale (i.e. never, sometimes and often) and higher scores indicate greater challenges. The BRIEF2 provides age and sex standardised T -scores ($M = 50$, $SD = 10$) for scale scores on the skills mentioned and index scores of BRI, ERI, and CRI and a Global Executive Composite (GEC). BRI includes Inhibit and Self-Monitor, ERI consists of Shift and Emotional Control, and finally, CRI encompasses Initiate, Working Memory, Plan/Organize, Task-Monitor, and Organisation of Materials. The manual reports test–retest reliability estimates for the nine scales ranging from 0.67 to 0.92, with a mean of 0.79 over an average time interval of 2.9 weeks (Gioia *et al.* 2015). Test–retest reliability of the BRIEF2 has been determined to be appropriate for use in DS (Esbensen *et al.* 2019), and the measure is suggested for use in children and adolescents with DS (Esbensen *et al.* 2017).

Cognition

Kaufman Brief Intelligence Test, second edition (KBIT-2), (Kaufman 2004), *Leiter International Performance Scale, Third Edition (Leiter-3)*, (Roid & Koch 2017), *Differential Ability Scales, Second Edition (DAS-II)*, (Elliott 2007), and *Stanford-Binet Intelligence Scales, Fifth Edition (SB5)*, (Roid 2003) estimated verbal and/or nonverbal cognitive abilities of the children and youth with DS. These measures are considered appropriate to describe overall intellectual ability in this population (Esbensen *et al.* 2017), and were used for descriptive purposes only in the current study.

Data analyses

Mean T -scores of the BRIEF2 indices were used for the analyses, and data were screened to detect any

univariate and multivariate outliers. No extreme univariate outliers (Z scores $<|3.29|$, (Field 2013)) were observed, and Mahalanobis distance considering the set of nine BRIEF2 clinical scales revealed that there were no multivariate outliers ($P > 0.001$). Descriptive and correlational analyses were performed to determine the mean, range, and score distribution for each scale and the correlations between scale pairs. Finally, a series of CFAs at the scale level were conducted in Mplus using maximum likelihood estimation to examine the factor structure of the BRIEF2 by investigating the fits for the pre-specified latent variable structures. These include one-, two-, and three-factor models proposed by the original developers of the BRIEF and BRIEF2 (Gioia 2000; Gioia *et al.* 2015; Gioia & Isquith 2002).

In the first model (one-factor model), all nine scales of the BRIEF2 were constrained to load on a single latent factor. The second model was a correlated two-factor model with four scales (Inhibit, Shift, Self-Monitor, and Emotional Control) loading on the first latent factor (BRI/ERI) and five scales (Working Memory, Initiate, Task-Monitor, Plan/Organize, and Organisation of Materials) loading on the second latent factor (CRI). In this model, Working Memory and Inhibit were the referent variables. Finally, the third model was a correlated three-factor model akin to the theoretical structure of the BRIEF2 with Working Memory, Inhibit, and Shift as the referent variables. No additional parameter constraints were added to the models. Bentler's comparative fit index (CFI), the Tucker–Lewis Index (TLI), the root mean square error of approximation (RMSEA), the standardised root mean square residual (SRMR), and Akaike information criteria (AIC) were chosen as goodness-of-fit indices. In addition, the Chi-Square difference test and AIC were used to compare fits between models (Keith 2019). The cut-off criteria for good fitting models were $RMSEA \leq 0.05$, $CFI \geq 0.95$, $TLI \geq 0.95$, $SRMR \leq 0.08$, and a smaller value of AIC (Keith 2019).

Results

Descriptive and correlation analyses

Mean, standard deviation, range, skewness, and kurtosis of the nine scales of the BRIEF2 were examined (Table 1). On average all but three ?

>subscales had mean T -scores within the normal range (<1 SD), and no subscale mean T -score reached the level of clinical significance ($T > 70$). The highest mean T -score was observed for Shift and the lowest mean T -score for Organisation of Materials. Skewness and kurtosis were between $+1$ and -1 indicating a normal distribution of T -scores. Pearson correlation coefficients between subscale T -scores ranged from 0.28 to 0.67 (Table 2). All were statistically significant when applying Bonferroni correction for multiple comparisons (adjusted P -value <0.00139).

Confirmatory factor analyses

Fit indices of the three proposed models and the chi-square differences between the models are displayed in Table 3. The one-factor model revealed a generally poor fit, with χ^2 being statistically significant and all fit indices failing to meet predetermined cut-off criteria.

The two-factor model (Model 2) yielded an acceptable fit according to the SRMR index; however, TLI, CFI, and RMSEA values failed to meet predetermined acceptable cutoff criteria. Further, there was a significant χ^2 difference between one- and two-factor models ($\Delta\chi^2(1) = 172$, $P < 0.001$) and smaller values of AIC implying that the more constrained two-factor model potentially explained the data better than the one-factor model. Finally, the three-factor model showed a good fit to the data in terms of CFI, RMSEA, and SRMR. A statistically significant chi-square difference between the two- and three-factor models ($\Delta\chi^2(2) = 27$, $P < 0.001$) and a

Table 1 Descriptive statistics of the BRIEF2 scales T -scores

Scale	Mean	SD	Range	Skewness	Kurtosis
Inhibit	58.91	11.24	37–90	0.42	−0.23
Self-Monitor	61.77	10.25	39–80	−0.09	−0.56
Shift	64.33	12.49	39–90	0.07	−0.74
Emotional Control	54.39	10.62	40–84	0.66	−0.17
Initiate	61.62	10.07	38–90	0.10	−0.27
Working Memory	63.24	9.05	38–89	0.05	−0.11
Plan/Organize	58.51	9.84	37–80	−0.06	−0.29
Task-Monitor	62.48	9.00	35–82	−0.59	0.24
Organisation of Materials	54.15	9.03	38–88	0.64	0.25

$N = 407$. SD, standard deviation.

Table 2 Correlation matrix of the BRIEF2 scales *T*-scores

	1	2	3	4	5	6	7	8
1. Inhibit	-							
2. Self-Monitor	0.52*	-						
3. Shift	0.58*	0.49*	-					
4. Emotional Control	0.65*	0.41*	0.63*	-				
5. Initiate	0.46*	0.48*	0.48*	0.30*	-			
6. Working Memory	0.55*	0.53*	0.49*	0.40*	0.67*	-		
7. Plan/Organize	0.47*	0.42*	0.46*	0.35*	0.62*	0.52*	-	
8. Task-Monitor	0.44*	0.44*	0.36*	0.28*	0.61*	0.60*	0.59*	-
9. Organisation of Materials	0.45*	0.37*	0.36*	0.32*	0.52*	0.57*	0.59*	0.51*

* $P < 0.00139$ (Bonferroni correction).

smaller value of AIC indicated that the less parsimonious three-factor model provided a better fit than the two-factor model.

See Figure 1 for a diagram of the best fitting, three-factor model. The diagram includes all standardised factor loadings, factor correlations, and residual variances with standard errors. All factor loadings exceeded 0.65, supporting the hypothesised correlation between the BRIEF2 scales and the three-factor structure (Gioia & Isquith 2002). There was a very high correlation ($r = 0.94$) between BRI and ERI, and lower correlations between these two indices (BRI and ERI) and the CRI ($r = 0.82$ and $r = 0.64$, respectively).

Discussion

Given the frequent use of the BRIEF2 for measuring EF in individuals with DS, there is a need to evaluate its factor structure in the DS population. The purpose of the present study was to examine the latent dimensionality of the BRIEF2 in a sample of youth

with DS through a series of CFAs to provide a useful window to understand how EF is structured uniquely within this particular clinical sample. Three competing models proposed by previous studies were analysed, including a three-factor correlated model akin to the theoretical factor structure of the BRIEF2, a two-factor correlated model, and a single-factor model. The results revealed that the three-factor model provided a better fit than the one- and two-factor models, yet most model fit indices were moderate.

Unitary versus dissociable

The findings support the dissociability theory of the EF construct among individuals with DS as both two- and three-factor models had significantly better fit than the one-factor model. An increasing emphasis has been placed on understanding the nature of EF as a unitary or non-unitary construct using different proxy-reported, self-reported and performance-based measures. Some scientists consider EF to be a single

Table 3 Fit indices and chi-square differences of the models

Factor model	χ^2	df	<i>P</i>	$\Delta\chi^2$	Δ df	<i>P</i>	AIC	RMSEA (90% CI)	CFI	TLI	SRMR
One-factor model	315	27	<0.001	-	-	-	25 818.3	0.16 (0.15, 0.18)	0.84	0.79	0.07
Two-factor model	143	26	<0.001	172	1	<0.001	25 648.3	0.10 (0.09, 0.12)	0.94	0.91	0.05
Three-factor model	116	24	<0.001	27	2	<0.001	25 625.5	0.09 (0.08, 0.11)	0.95	0.93	0.04

df, model degrees of freedom; CI, confidence interval; SRMSR, standardised root mean squared residual; AIC, Akaike information criteria; RMSEA, root means squared error of approximation; TLI, Tucker–Lewis Index, CFI, comparative fit index.

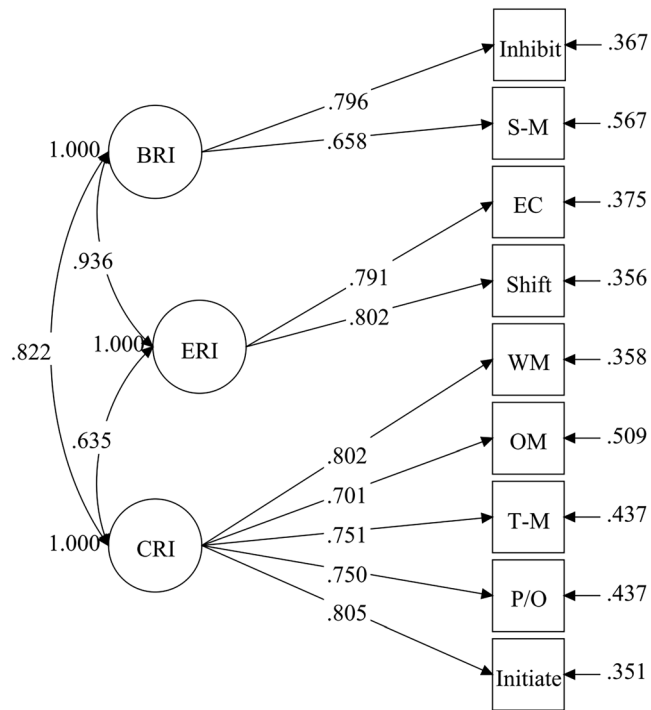


Figure 1. Diagram of the three-factor model. BRI, Behavioral Regulation Index; ERI, Emotional Regulation Index; CRI, Cognitive Regulation Index; S-M, Self-Monitor; EC, Emotional Control; WM, Working Memory; P/O, Plan/Organize; T-M, Task-Monitor; OM, Organisation of Materials.

entity in the early stages of the development in typically developing children without including any distinct subfunctions or subcomponents (Duncan *et al.* 1996; Duncan *et al.* 1997; Teuber 1972). Further, from a developmental perspective, some researchers have shown that EF is not separable in younger children, but does indeed become separable yet related at about the age of 12 years in typical development (Laureys *et al.* 2022; Xu *et al.* 2013). Behavioural and genetic evidence suggests that EF is characterised by relatively independent but related subcomponents specifically in typically developing participants in older ages (for a comprehensive review see (Friedman & Miyake 2017, Karr *et al.* 2018). In addition, functional neuroimaging data support the existence of partially separable but overlapping EF processes at the neural level in typically developing children over 6 years (McKenna *et al.* 2017). Specifically, the non-unitary nature of the EF construct, as measured by BRIEF and BRIEF2, has been supported in the DS population (Lee *et al.* 2015; Loveall *et al.* 2017). In line with most relevant literature, the results of the current study provide further evidence for differentiation of EF as measured by the BRIEF2, a proxy-reported

measure, in a sample of youth with DS ages 6–18 years.

Two-factor model versus three-factor model

Based on the model fitting criteria, the results of the current study provided some evidence to support the theoretical structure of the BRIEF2 in which CRI, BRI, and ERI are conceptualised as distinct dimensions (Gioia *et al.* 2015), although a close connection between behavioural and emotional regulation factors was observed in the model ($r = 0.94$). A large correlation between BRI and ERI has also been reported in previous studies documenting support for the three-factor model of both BRIEF and BRIEF2 in different clinical samples. For example, Gioia and Isquith (2002) found a large overlap ($r = 0.84$) between behavioural and emotional factors of the BRIEF in a mixed clinical sample. Similarly, a correlation of 0.83 was reported between behavioural and emotional factors of the BRIEF in a mixed healthy and clinical sample (Egeland & Fallmyr 2010). Further, a large correlation between BRI and ERI was also found in a study that supported the three-factor model of the

BRIEF2 in children with ADHD (Parhoon *et al.* 2022). Thus, separating BRI from ERI and conceptualising them as distinct components may provide meaningful explanatory value for the BRIEF2 among youth with DS.

Alternatively, It is possible that dysregulated behavioural and emotional functioning are less differentiable on parent reports in clinical samples (Lace *et al.* 2021) including those with DS, and the two-factor model with the merged BRI and ERI makes sense. In the current study, we selected the three-factor structure as the best fitting model based on chi-square differences and its relevant *P*-value. However, chi-square differences can be affected by sample size. Compared with other studies that supported the two-factor model of the BRIEF and BRIEF2 ((Lace *et al.* 2021, Lyons Usher *et al.* 2016, Slick *et al.* 2006), the relatively larger sample size in the current study can result in higher chi-square differences between the models and consequently increase the chance to have a significant *P*-value. Thus, regarding small differences in other fit indices between the two- and three-factor models, and also the large correlation between BRI and ERI in the three-factor model, it is possible that the two-factor model theoretically explains the underlying structure of the BRIEF2 in this sample of youth with DS.

Limitations and future directions

While the present study evaluated and compared the three competing models of the BRIEF2 in a sample of children and youth with DS, some limitations need to be acknowledged. First, due to the confirmatory nature of the study, we did not impose any error correlation or cross-loadings to improve the model fits. Further research is recommended to conduct exploratory factor analyses to determine how the items may load on subscales and latent profile analyses to determine possible subgroups with differing EF profiles in DS. Second, to avoid Heywood cases, we did not impose additional parameters on the models. Future directions are suggested to examine more complex models proposed by previous research such as higher-order and/or bifactor models in samples of participants with DS. Third, IQ measures were unavailable for several participants, and we were unable to examine the effect of IQ on the factor structure of the BRIEF2 in this

sample of youth with DS. Further research is needed to carry out measurement invariance analysis across IQ levels and examine the stability of fit indices among participants' subgroups. Fourth, the sample lacked diversity in race and ethnicity and future recruitment efforts to expand on diversity are warranted. Finally, because we analysed BRIEF2 *T*-scores that were age normed, we did not conduct measurement invariance analysis across age, and ongoing study is recommended to carry out such analysis using BRIEF2 raw scores.

Conclusions

This study addressed a notable gap in the literature regarding the structure of the BRIEF2 in youth with DS. Results suggest interpretation of EF indices in terms of behavioural regulation, emotional regulation, and cognitive regulation. The better fitting three-factor model informs clinical practice and supports the ongoing validation of outcome measures for use in research studies and clinical trials with young people with DS.

Acknowledgements

This research would not have been possible without the contributions of the participating families and the community support.

Source of funding

This manuscript was prepared with support from the Eunice Kennedy Shriver National Institute of Child Health and Human Development of the National Institutes of Health (P50 HD103526 Abbeduto; R03 HD083596 Channell; R01 HD093754 Esbensen; R61 HD100934 Esbensen/Froehlich; R01 HD099150 Fidler; R21 HD101000 Fidler; KL2TR002490 Grzadzinski; R01 HD055741 Hazlett; R21 HD100997 Lee; R21 HD106164 Lee; U54 HD079123 Schlaggar, T32 HD007489 Hartley), National Center for Advancing Translational Science: KL2TR002490, the Lejeune Foundation (1447 Lee), and the Jack Rubinstein Foundation for Developmental Disabilities. The content is solely the responsibility of the authors and does not necessarily represent the official views of the National Institutes of Health.

Conflict of interest

The authors have no conflicts of interest to disclose.

Data availability statement

Data are available from the last author upon request. Data from the current study were combined from six studies, and all study procedures were approved by their respective site Institutional Review Boards (IRBs). Data sharing or data use agreements were also obtained. Parental consent to participate was obtained, and child assent as appropriate per site IRB requirements.

References

- Amadó A., Serrat E. & Vallès-Majoral E. (2016) The role of executive functions in social cognition among children with down syndrome: relationship patterns. *Frontiers in Psychology* **7**, 1363.
- Daunhauer L. A., Fidler D. J., Hahn L., Will E., Lee N. R. & Hepburn S. (2014) Profiles of everyday executive functioning in young children with Down syndrome. *American Journal on Intellectual and Developmental Disabilities* **119**, 303–18.
- Daunhauer L. A., Gerlach-McDonald B., Will E. & Fidler D. (2017) Performance and ratings based measures of executive function in school-aged children with Down syndrome. *Developmental Neuropsychology* **42**, 351–68.
- Daunhauer L. A., Will E., Schworer E. K. & Fidler D. J. (2020) Young students with Down syndrome: Early longitudinal academic achievement and neuropsychological predictors. *Journal of Intellectual and Developmental Disability* **45**, 211–21.
- Diamond A. (2013) Executive functions. *Annual Review of Psychology* **64**, 135–68.
- Dodzik P. (2017) Behavior Rating Inventory of Executive Function, Gerard A. Gioia, Peter K. Isquith, Steven C. Guy, and Lauren Kenworthy. *Journal of Pediatric Neuropsychology* **3**, 227–31.
- Duncan J., Emslie H., Williams P., Johnson R. & Freer C. (1996) Intelligence and the frontal lobe: The organization of goal-directed behavior. *Cognitive Psychology* **30**, 257–303.
- Duncan R., Johnson M., Swales C. & Freer J. (1997) Frontal lobe deficits after head injury: Unity and diversity of function. *Cognitive Neuropsychology* **14**, 713–41.
- Egeland J. & Fallmyr Ø. (2010) Confirmatory factor analysis of the Behavior Rating Inventory of Executive Function (BRIEF): Support for a distinction between emotional and behavioral regulation. *Child Neuropsychology* **16**, 326–37.
- Elliott C. D. (2007) *Differential ability scales, (DAS-II)*, Trans. Pearson, San Antonio, TX.
- Esbensen A. J., Hoffman E., Shaffer R., Chen E., Patel L. & Jacola L. M. (2019) Reliability of informant report measure of executive functioning in children with Down syndrome. *American Journal of Intellectual and Developmental Disabilities* **124**, 220–33.
- Esbensen A. J., Hoffman E. K., Shaffer R. C., Patel L. R. & Jacola L. M. (2021) Relationship Between Parent and Teacher Reported Executive Functioning and Maladaptive Behaviors in Children With Down Syndrome. *American Journal on Intellectual and Developmental Disabilities* **126**, 307–23.
- Esbensen A. J., Hooper S. R., Fidler D., Hartley S. L., Edgin J. O., d'Ardhuy X. L. *et al.* (2017) Outcome measures for clinical trials in Down syndrome. *American Journal on Intellectual and Developmental Disabilities* **122**, 247–81.
- Esbensen A. J. & MacLean W. (2017) Down syndrome. In: *A Comprehensive Guide to Intellectual and Developmental Disabilities* (eds M. Wehmeyer, K. Shogren, A. Fung, M. Percy & I. Brown), pp. 195–208. Brookes, Baltimore, MD.
- Field A. (2013) *Discovering statistics using IBM SPSS statistics*, Trans. Sage.
- Friedman N. P. & Miyake A. (2017) Unity and diversity of executive functions: Individual differences as a window on cognitive structure. *Cortex* **86**, 186–204.
- Gioia G. A. (2000) *Behavior Rating Inventory of Executive Function: Professional Manual*, Trans. Psychological Assessment Resources, Incorporated, Odessa, FL.
- Gioia G. & Isquith P. (2002) Two faces of monitor: They self and thy task. *Journal of the International Neuropsychological Society* **8**, 229.
- Gioia G. A., Isquith P. K., Guy S. C. & Kenworthy L. (2015) *Behavior Rating Inventory of Executive Function 2nd Edition (BRIEF2): Professional Manual*, Trans. Psychological Assessment Resources, Incorporated, Odessa, FL.
- Hendrickson, N. K., & McCrimmon, A. W. (2018). Test review: behavior rating inventory of executive function®, second edition (BRIEF®2) by Gioia, G. A., Isquith, P. K., Guy, S. C., & Kenworthy, L. *Canadian Journal of School Psychology*, **34**, 73–8.
- Iralde L., Roy A., Detroy J. & Allain P. (2020) A Representational Approach to Executive Function Impairments in Young Adults with Down Syndrome. *Developmental Neuropsychology* **45**, 263–78.
- Jacobson L. A., Kalb L. G. & Mahone E. M. (2020) When theory met data: Factor structure of the BRIEF2 in a clinical sample. *The Clinical Neuropsychologist* **34**, 243–58.
- Karr J. E., Areshenkoff C. N., Rast P., Hofer S. M., Iverson G. L. & Garcia-Barrera M. A. (2018) The unity and diversity of executive functions: A systematic review and re-analysis of latent variable studies. *Psychological Bulletin* **144**, 1147.
- Kaufman A. (2004) *KBIT-2: Kaufman Brief Intelligence Test*, Second edn. Pearson, Upper Saddle River, NJ.

- Keith T. Z. (2019) *Multiple regression and beyond: An introduction to multiple regression and structural equation modeling*, Trans. Routledge.
- Kristensen, K., Lorenz, K. M., Zhou, X., Piro-Gambetti, B., Hartley, S. L., Godar, S. P., *et al.* (2022). Language and executive functioning in young adults with Down syndrome. *Journal of Intellectual Disability Research* **66**, 151–61.
- Lace J. W., Emmert N. A., Merz Z. C., Zane K. L., Grant A. F., Aylward S. *et al.* (2019) Investigating the BRIEF and BRIEF-SR in adolescents with mild traumatic brain injury. *Journal of Pediatric Neuropsychology* **5**, 9–19.
- Lace J. W., Seitz D. J., Austin T. A., Kennedy E. E., Ferguson B. J. & Mohrland M. D. (2021) The dimensionality of the Behavior Rating Inventory of Executive Function. In: *A clinical sample*, pp. 1–12. Applied Neuropsychology: Child.
- Lanfranchi S., Jerman O., Dal Pont E., Alberti A. & Vianello R. (2010) Executive function in adolescents with Down Syndrome. *Journal of Intellectual Disability Research* **54**, 308–19.
- Laureys F., De Waelle S., Barendse M. T., Lenoir M. & Deconinck F. J. (2022) The factor structure of executive function in childhood and adolescence. *Intelligence* **90**, 101600.
- Lee N. R., Anand P., Will E., Adeyemi E. I., Clasen L. S., Blumenthal J. D. *et al.* (2015) Everyday executive functions in Down syndrome from early childhood to young adulthood: evidence for both unique and shared characteristics compared to youth with sex chromosome trisomy (XXX and XXY). *Frontiers in Behavioral Neuroscience* **9**, 264.
- Lee N. R., Fidler D. J., Blakeley-Smith A., Daunhauer L., Robinson C. & Hepburn S. L. (2011) Caregiver report of executive functioning in a population-based sample of young children with Down syndrome. *American Journal on Intellectual and Developmental Disabilities* **116**, 290–304.
- Llewellyn C., Ayers S., McManus I., Newman S. P., Petrie K., Revenson T. *et al.* (2019) *Cambridge handbook of psychology, health and medicine*, Trans. Cambridge University Press.
- Loveall S., Conners F., Tungate A., Hahn L. & Osso T. (2017) A cross-sectional analysis of executive function in Down syndrome from 2 to 35 years. *Journal of Intellectual Disability Research* **61**, 877–87.
- Lyons Usher A. M., Leon S. C., Stanford L. D., Holmbeck G. N. & Bryant F. B. (2016) Confirmatory factor analysis of the Behavior Rating Inventory of Executive Functioning (BRIEF) in children and adolescents with ADHD. *Child Neuropsychology* **22**, 907–18.
- Mai C. T., Isenburg J. L., Canfield M. A., Meyer R. E., Correa A., Alverson C. J. *et al.* (2019) National population-based estimates for major birth defects, 2010–2014. *Birth Defects Research* **111**, 1420–35.
- Maiman M., Hamner T., Udhmani M., Anand P., Olesen L. S., Giedd J. N. *et al.* (2017) Differential effects of sex and intellectual ability level on "cool" and "hot" executive function impairments in youth with Down syndrome. In: *Paper Presented at the Gatlinburg Conference on Research and Theory in Intellectual and Developmental Disabilities*, San Antonio, TX.
- Manrique-Niño J., Díaz-Forero A., Velez-van Meerbeke A., Ramírez-Guerrero S., Florez-Esparza G. & Talero-Gutiérrez C. (2020) Executive function in down syndrome children in Bogotá, Colombia. *Heliyon* **6**, e05585.
- McKenna R., Rushe T. & Woodcock K. A. (2017) Informing the structure of executive function in children: a meta-analysis of functional neuroimaging data. *Frontiers in Human Neuroscience* **11**, 154.
- Memisevic H. (2015) Factor structure of behavior rating inventory of executive functions in children with intellectual disability. *Acta Neuropsychologica* **13**, 137–44.
- Ozonoff S. & Jensen J. (1999) Brief report: Specific executive function profiles in three neurodevelopmental disorders. *Journal of Autism and Developmental Disorders* **29**, 171–7.
- Parhoon K., Moradi A., Alizadeh H., Parhoon H., Sadaphal D. P. & Coolidge F. L. (2022) Psychometric properties of the behavior rating inventory of executive function, (BRIEF2) in a sample of children with ADHD in Iran. *Child Neuropsychology* **28**, 427–36.
- Roid G. H. (2003) *Stanford-binet intelligence scales (SB5)*, Trans. Riverside.
- Roid G. H. & Koch C. (2017) *Leiter-3: Nonverbal Cognitive and Neuropsychological Assessment*. In: *Handbook of Nonverbal Assessment* (ed. R. S. McCallum), pp. 127–50. Springer, New York, NY.
- Sabat C., Arango P., Tassé M. J. & Tenorio M. (2020) Different abilities needed at home and school: The relation between executive function and adaptive behaviour in adolescents with Down syndrome. *Scientific Reports* **10**, 1–10.
- Schworer E. K., Fidler D. J., Kaur M., Needham A., Prince M. A. & Daunhauer L. A. (2022) Infant precursors of executive function in Down syndrome. *Journal of Intellectual Disability Research*, 108–20.
- Slick D. J., Lautzenhiser A., Sherman E. M. & Eylr K. (2006) Frequency of scale elevations and factor structure of the Behavior Rating Inventory of Executive Function (BRIEF) in children and adolescents with intractable epilepsy. *Child Neuropsychology* **12**, 181–9.
- Teuber H.-L. (1972) Unity and Diversity of Frontal Lobe Functions. *Acta Neurobiologiae Experimentalis* **32**, 615–56.
- Tomaszewski B., Fidler D., Talapatra D. & Riley K. (2018) Adaptive behaviour, executive function and employment in adults with Down syndrome. *Journal of Intellectual Disability Research* **62**, 41–52.

- Tungate A. S. & Conners F. A. (2021) Executive function in Down syndrome: A meta-analysis. *Research in Developmental Disabilities* **108**, 103802.
- de Weger C., Boonstra F. N. & Goossens J. (2021) Differences between children with Down syndrome and typically developing children in adaptive behaviour, executive functions and visual acuity. *Scientific Reports* **11**, 1–15.
- Will E., Fidler D., Daunhauer L. & Gerlach-McDonald B. (2017) Executive function and academic achievement in primary-grade students with Down syndrome. *Journal of Intellectual Disability Research* **61**, 181–95.
- Will E. A., Schworer E. K. & Esbensen A. J. (2021) The role of distinct executive functions on adaptive behavior in children and adolescents with Down syndrome. *Child Neuropsychology*, 1–19.
- Xu F., Han Y., Sabbagh M. A., Wang T., Ren X. & Li C. (2013) Developmental differences in the structure of executive function in middle childhood and adolescence. *PLoS ONE* **8**, e77770.

Accepted 28 November 2022