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**Special Issue Article**

# Profiles and trajectories of executive functioning in young children with Down syndrome

**A. Dimachkie Nunnally,[1] L. Baczewski,[2] K. Sterrett,[2] A. Holbrook,[2] A. Kaiser[3] & C. Kasari[2]**

1 *M.I.N.D Institute, University of California, Davis, CA, USA*
2 *Semel Institute for Neuroscience and Human Behavior, University of California, Los Angeles, CA, USA*
3 *Department of Special Education, Vanderbilt University, Nashville, TN, USA*

## Abstract

*Background* Language acquisition strongly predicts executive functioning (EF) in early childhood in typical development and in children with Down syndrome (DS). Both language and EF are critical contributors to later positive social and academic outcomes yet are often areas of concern in children with DS. Despite the wider availability of interventions targeting language development in DS, no efforts have been made to understand how these interventions may influence the development of EF in this population.
*Methods* This study examined secondary data from 76 preschoolers with DS collected as part of a randomised waitlist control trial of an early social communication intervention (JASPER-EMT). Children's EF skills were measured using the BRIEF-P, at three timepoints over 6 months. Linear regression was used to examine the baseline relationship between child characteristics and the three indices of the BRIEF-P: Emergent Metacognition, Flexibility and Inhibitory Self-Control. Linear mixed effects models were used to estimate change across the three indices of the BRIEF-P and whether that change was moderated by treatment.
*Results* Children in this sample exhibited an uneven profile of EF at baseline, with relative strengths in the Flexibility Index and the Inhibitory Self-Control Index, and relative weaknesses in the Emerging Metacognition Index. Chronological age was associated with all indices at baseline (all $P < 0.05$). Children in the intervention group exhibited improvements in the Flexibility Index from entry to exit (3 months later) compared with the control, although this treatment effect did not maintain at the follow up at 6 months.
*Conclusions* Baseline EF profiles of children were consistent with findings of other studies with children with DS. Longitudinal findings suggest that behavioural interventions targeting language may have positive collateral effects on certain EF skills, however these effects may be transitory without ongoing support. These findings illustrate both the need for further exploration of the impact of early language interventions on EF abilities and the malleability of certain EF domains in young children with DS.

**Keywords** Down syndrome, early Intervention, executive function, intellectual disability

Correspondence: Dr Amanda Dimachkie Nunnally, M.I.N.D Institute, University of California, 2825 50th Street, Suite 2101 Sacramento, Davis, CA 95817, USA (e-mail: adimachkie@ucdavis.edu).

## Introduction

Executive functioning (EF) refers to a set of higher-order cognitive skills that are responsible for purposeful, goal-directed behaviour (Gioia *et al.* 2001). Included among those skills are working memory, planning and organisational skills, inhibitory control, attentional shifting and emotion control (Brock *et al.* 2009; Willoughby *et al.* 2012; Sasser *et al.* 2015), all skills that are necessary for children's successful navigation of the growing cognitive and social demands of their environment (Miyake *et al.* 2002; Razza 2009). Children with Down syndrome (DS) display a unique EF profile characterised by areas of relative strengths and weaknesses (Daunhauer *et al.* 2014), which are related to delays in expressive and receptive language that are characteristic of DS (Grieco *et al.* 2015). Although these two developmental domains are clearly intertwined, there has been little work examining their relationship across early development in children with DS.

### EF in early childhood

During the first 5 years of life, rapid brain maturation leads to the development of diverse cognitive skills, including EF (Bell & Fox 1992; Klingberg *et al.* 1999; Best & Miller 2010). The acquisition of EF skills in early life is related to the development of language for both TD children as well as children with DS (Kuhn *et al.* 2014; Petersen *et al.* 2015). In one study, TD children's growth in expressive language abilities between 24 and 36 months of age was predictive of their EF skills at 60 months of age (Kuhn *et al.* 2016). Interestingly, expressive language abilities after 36 months of age no longer predicted EF abilities at 60 months of age, suggesting an important role played by early language acquisition in the development of EF skills. Some researchers have hypothesized that this relationship can be explained by the notion that language acquisition is necessary for abstract and representational thinking, processes that are fundamental to many EF tasks (Zelazo & Frye 1998; Zelazo *et al.* 2003). Although there is still much to learn about the relationship between language acquisition and EF in early childhood, it is apparent that these domains are linked (Grieco *et al.* 2015).

### EF and language in children with DS

DS is the leading genetic cause of intellectual disability (ID), with recent prevalence rates of 1 in 700 births (Mai *et al.* 2019). Most often, DS results from the presence of a third copy of all or part of chromosome 21, resulting in a characteristic phenotype marked by delays across cognitive domains (Antonarakis & Epstein 2006), including language development and EF abilities (Miller 1995; Gioia *et al.* 2000; Lanfranchi *et al.* 2010).

Studies have consistently documented challenges in EF for children with DS, primarily in the domains of working memory and planning and organisational skills (Daunhauer *et al.* 2014; Loveall *et al.* 2017). In contrast, children with DS exhibit relative strengths in emotion control, performing similarly to their TD peers (Lee *et al.* 2011). Mixed findings exist regarding performance in other EF domains, such as inhibitory control and attentional shifting (Lee *et al.* 2011; Carney *et al.* 2013; Daunhauer *et al.* 2014); however, these inconsistencies may be in part due to differences in the type of tasks used, reporter and age. Notably, a cross sectional analysis of EF in children with DS found that while younger children exhibited a relative strength in attentional shifting, challenges in this domain emerged among older children in this population (Loveall *et al.* 2017). These findings suggest that children with DS do not keep pace with peers in the development of these skills over time. Overall, these findings illustrate an asynchronous pattern of abilities and challenges across EF domains among older children with DS, possibly attributable to delays in development, although they have not yet been widely replicated using a sample of younger children with DS.

Children with DS also exhibit delays in both expressive and receptive language (Abbeduto *et al.* 2006). Given findings that early expressive language predicts later EF abilities in TD children (Kuhn *et al.* 2016) and the association between expressive language and EF skills in older children with DS (Campbell *et al.* 2013; Udhnani *et al.* 2020), it is essential to explore whether social communication interventions for children with DS may impact the development of EF.

### The developmental approach

Ed Zigler was one of the most influential figures in the field of disability research in the 20th century, and his contributions to science have continued to shape the way that researchers and clinicians think about individuals with intellectual disability (ID). Of note, Ed Zigler was a champion of the universal developmental approach, which espouses the belief that development is a universally human experience, regardless of ID status (Burack *et al.* 2021; Hodapp 2021). His approach emphasised that individuals with ID follow similar developmental trajectories as do their typical developing (TD) peers, albeit at a slower pace. For individuals with ID of known aetiology, such as those with DS, this development may occur in an asynchronous fashion, such that uneven profiles of development may occur. Consistent with the belief that development is a universal human experience, Zigler emphasised the important contribution of context, such as intervention, in shaping development for children with ID and TD alike. By highlighting the shared universal experience of development, Ed's work contributed to efforts to humanise and de-stigmatise individuals with ID, who have been historically marginalised and stigmatised for centuries Additionally, this perspective has influenced the way that researchers and clinicians regard development in individuals with ID, by giving consideration to the different ways that development can be supported for these individuals. This paper aims to leverage these perspectives to explore the development of EF abilities in children with DS, an ID with known aetiology. First, we approach the development of EF in children with DS as comparable to their TD peers, suggesting that the development of EF in children with DS relies on the same developmental processes as in TD, albeit with delays. We also explore the idea that the development of EF in children with DS may follow an asynchronous pattern, exhibiting an uneven profile of strengths and weaknesses. Lastly, we consider the impact of context, more specifically, a parent-mediated language intervention, on the way in which EF abilities develop in young children with DS.

Thus, the present study aimed to (1) characterise the early EF profile in a sample of ~~very~~ language delayed preschool children with DS as measured by a parent report of EF (the BRIEF-P) and (2) evaluate whether a naturalistic developmental behavioural intervention targeting early social communication (JASPER-EMT) improves EF skills, over time in a sample of young children with DS.

Consistent with extant studies that examine EF using the BRIEF-P, we centre hypotheses around the three main indices: Flexibility (FI), Emergent Metacognition (EMI) and Inhibitory Self-Control (ISCI). Based on the sparse literature examining EF in young children with DS, we hypothesized that children would exhibit relative strengths in emotion control and attentional shifting, as measured by the Flexibility Index, and relative weaknesses in working memory, as measured by the Emerging Metacognition Index, at baseline. Furthermore, at baseline, higher expressive and receptive language abilities would be related to higher EF skills across domains. It was also hypothesized that children who were assigned to receive JASPER-EMT would exhibit relative improvements in EF abilities, based on the targeted focus of the intervention on early social communication.

## Methods

### Participants

Participants included 76 children with DS ($M_{\text{BaselineAge}}$ = 38.99 months, *SD* = 7.17) drawn from a randomised controlled intervention trial of an early social communication intervention (JASPER-Enhanced Milieu Teaching; JASPER-EMT) conducted at two sites. JASPER-EMT is an established intervention approach that has been tested in populations of young children with other neurodevelopmental disorders such as autism (Kasari *et al.* 2014). This approach combines principles from two evidence-based, naturalistic developmental behavioural interventions (Joint Attention, Symbolic Play, Engagement & Regulation; JASPER; Kasari *et al.* 2008, 2010) and Enhanced Milieu Teaching; EMT (Kaiser & Roberts 2011) to produce a blended treatment approach that targets the unique areas of strength and weaknesses of the DS population. Families were recruited via advertisements in community centres, schools, and Down syndrome associations in each city. Children were eligible to participate if they had a diagnosis of DS and (1) were 30–54 months at study entry, (2) had less than 20 functional words (calculated using PLS-5 in either English and/or Spanish) and (3) tested at the 18-month mental age level or above on the visual reception scale of the Mullen Scales of Early Learning

(MSEL). Children with comorbid diagnoses (e.g. DS and autism) were excluded from the study.

## Procedures

Children who met inclusion criteria were randomly assigned to the immediate treatment ($n$ = 38) or waitlist control groups ($n$ = 40) (Figure 1). Children assigned to the waitlist control group did not receive intervention during the active period of the study, instead receiving intervention after the 6-month follow up. Although bilingual families were not specifically recruited for the study, approximately half of the full sample, and 82% of children in the immediate intervention group, were reported to be bilingual by their parents. Bilingualism was determined via a caregiver-report question that was included in the demographic form. Caregivers were asked whether their family spoke another language in the home, in addition to English. Consistent with the ethnic and racial demographics of the two study sites, 22% of families at Site 1 were reported to be bilingual, while 82.5% of families at Site 2 reported to be bilingual (majority in Spanish and English).

Participants' EF abilities were assessed using the BRIEF-P at three study time points: Time 1 (T1; prior to intervention), Time 2 (T2; post-intervention at the 3-month follow-up), and Time 3 (T3; 6-month follow-up). Assessments were conducted by graduate student or staff researchers blind to treatment condition. Caregivers completed questionnaires about demographic characteristics at T1 and T3.

## Measures

### *Behaviour Rating of Executive Function - Preschool Version (BRIEF-P)*

The Behaviour Rating of Executive Function - Preschool Version (BRIEF-P; Gioia *et al.* 2003) was used to measure EF. The BRIEF-P is a standardised 63-item caregiver/teacher-report questionnaire designed to measure EF skills among preschool-aged children. Although the BRIEF-P was normed for use among neurotypical children (Gioia *et al.* 2003) it has been extensively used to measure EF skills among children with neurodevelopmental diagnoses, including children with DS (Lee *et al.* 2011; Daunhauer *et al.* 2014; Loveall *et al.* 2017).

The measure consists of five subdomains that each represent an aspect of EF: emotional control, working memory, inhibit, shifting, and planning/organising. These subdomains are grouped into three indices: Flexibility Index (FI), Emergent Metacognition Index (EMI), and Inhibitory Self-Control Index (ISCI). $T$-scores ($M$ = 50, $SD$ = 10) are generated for each index, such that $T$-scores of 65 or above (1.5 SDs above the normative mean) on each index are considered 'abnormally elevated' and clinically significant. For each item, the caregiver is asked to rate how often, during the last 6 months, the behaviour has been a concern. Items are measured on

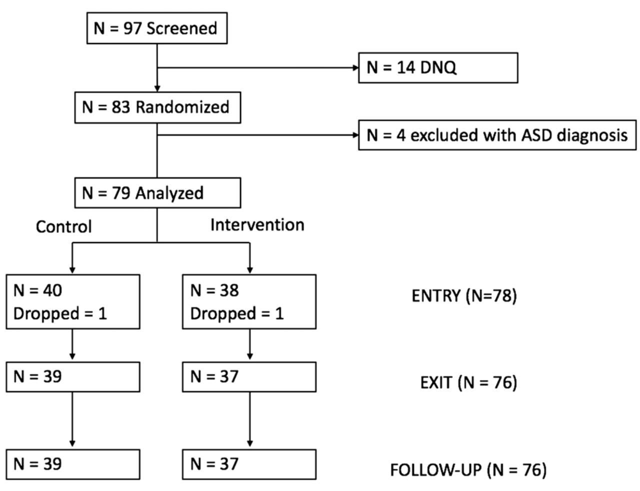


**Figure 1.** Consort diagram.

a three-point Likert scale (1 = never, 2 = sometimes, 3 = often). For the current study, caregivers completed the measure in either English or Spanish at T1, T2 and T3.

*Flexibility Index (FI).* This index measures the child's ability to flexibly shift their attention between tasks, parts of a problem, or situations. Additionally, this index includes items that measure emotional control, or the child's ability to modulate their emotions and corresponding behavioural responses.

*Emergent Metacognition Index (EMI).* Items on this index measure working memory, planning and organisation skills. Working memory refers to one's ability to maintain and hold information in their attentional focus in order to complete a task or generate an appropriate response. Planning and organisational skills include the ability to anticipate and prepare for future activities, tasks, or demands.

*Inhibitory Self-Control Index (ISCI).* Items measuring the child's inhibition and emotional control make up the inhibitory self-control index. Inhibition refers to one's ability to inhibit responses and avoid engaging in impulsive or inappropriate behaviour.

### *Preschool language scales, 5th edition (PLS-5)*

The Preschool Language Scales, 5th Edition (PLS-5; Zimmerman *et al.* 2011), is a standardised developmental language assessment used with children ages birth to 7 years, 11 months old. The PLS-5 has been used to measure expressive and receptive language in studies of children with developmental delay, autism, and language impairment (Zimmerman *et al.* 2011). The present study utilised age equivalent scores from Auditory Comprehension and Expressive Communication subscales of the PLS-5 at both T1 and T2. Each subscale generates a standard score, percentile rank, and age equivalent score. The PLS-5 is available in both English and Spanish, children whose primary language was Spanish were administered the Spanish version.

### *Mullen Scales of Early Learning (MSEL)*

The MSEL is a standardised developmental assessment that evaluates verbal and nonverbal abilities of children between the ages of 1–68 months old (Mullen 1995). A trained assessor presents the child with a series of tasks and questions grouped into five subdomains: (1) gross motor, (2) fine motor, (3) visual reception, (4) receptive language and (5) expressive language. Analyses for this study used visual reception age equivalent scores.

### *JASPER-EMT*

JASPER-EMT is a blended intervention that incorporates elements of two evidence-based, naturalistic developmental behavioural interventions, JASPER and Enhanced Milieu Teaching (EMT), designed to target early social-communication in children with DS. This blended intervention has been designed in order to foster engagement, teach play and promote children's language development.

### *JASPER*

A naturalistic, play-based behavioural intervention, JASPER focuses on children's spontaneous initiations of social communication gestures, language and play acts in order to improve children's initiation of social communication skills (Kasari *et al.* 2006, 2014).

*Enhanced Milieu Teaching (EMT).* EMT is a naturalistic early language intervention that capitalises on children's initiations and interests as opportunities to prompt and model language in day-to-day contexts (Kaiser 1993; Kaiser & Hampton 2017). It combines behavioural teaching strategies with developmentally appropriate interaction strategies (responsiveness, modelling target language and expanding children's utterances) in order to increase the frequency and complexity of children's language.

Participants assigned to the JASPER-EMT group received one-on-one sessions twice a week with a trained therapist, in addition to two caregiver-coaching sessions per week, over the course of 12 weeks. One-on-one intervention sessions were delivered to the child either in the home or at the child's preschool, while caregiver-coaching sessions were administered at home. Due to the large number of bilingual families enrolled in the study, therapist intervention sessions were conducted in English and caregiver coaching was offered in English or Spanish, depending on the caregiver's preference.

## Data analysis

Means and standard deviations were calculated for each of the BRIEF-P indices separately. The outcome of the randomization was assessed across demographic and outcome variables using T-tests for continuous variables and chi-squared tests for categorical variables. One-way ANOVAs were used to compare the t-scores across the three indices at entry, significant ANOVAs were followed up with pairwise comparisons between each index and Tukey's correction was applied.

Next, separate linear regression models were fit to assess the relationship between each BRIEF-P index and child demographic characteristics. Each BRIEF-P index was modelled separately as a dependent variable, the independent variables in the models included PLS-5 expressive and auditory comprehension age equivalent scores, chronological age, and children's age equivalent visual receptive scores.

Lastly, to test the longitudinal aim, separate generalised linear mixed models (GLMM) were fit for each index. Fixed effects for each model included site, time (T1, T2, T3) and treatment (JASPER-EMT versus control) as categorical variables, as well as a time by treatment interaction term. We also tested a time by treatment by site interaction for each longitudinal model. Additional fixed effects included receptive language age equivalent scores and CA. Participants were entered into the models as random effects (intercept).

# Results

## Descriptive statistics

### *Children's characteristics at baseline*

Means and standard deviations for demographic information at baseline are presented in Table 1. On average, children in the sample had an Auditory

**Table 1** Caregiver and child characteristics

| Measure | Variable | Value | DS (*n* = 79) Mean (SD) or *N* (%) | Control (*n* = 41) Mean (SD) or *N* (%) | JASPER-EMT (*n* = 38) Mean (SD) or *N* (%) | *P* value |
|---|---|---|---|---|---|---|
| Demographic (Child) | Child's age | (months) | 38.99 (7.17) | 39.02 (7.21) | 38.95 (7.23) | 0.96 |
| | Gender | Male | 37 (47%) | 20 (49%) | 17 (45%) | 0.89 |
| | | Female | 42 (53%) | 21 (51%) | 21 (55%) | |
| | Child's race | White | 51 (70%) | 26 (68%) | 25 (71%) | 0.42 |
| | | Multiple | 9 (12%) | 5 (13%) | 4 (11%) | |
| | | Asian | 3 (4%) | 3 (8%) | 0 (0%) | |
| | | African American | 2 (3%) | 1 (3%) | 1 (3%) | |
| | | Other | 1 (1%) | 1 (3%) | 0 (0%) | |
| | | Did not respond | 7 (10%) | 2 (5%) | 5 (14%) | |
| | Child's ethnicity | Hispanic/Latino | 33 (45%) | 16 (42%) | 17 (47%) | 0.83 |
| | | Not Hispanic/Latino | 41 (55%) | 22 (58%) | 19 (53%) | |
| Mullen Scales of Early Learning | Visual receptive | (raw score) | 24.82 (3.87) | 25.23 (4.28) | 24.39 (3.41) | 0.35 |
| | | (standard score) | 22.87 (6.10) | 23.65 (6.99) | 22.05 (4.95) | 0.25 |
| | | (age equivalence months) | 22.51 (4.94) | 22.98 (5.37) | 22.03 (4.46) | 0.40 |
| | Fine motor | (raw score) | 22.48 (3.11) | 23.07 (3.05) | 21.84 (3.09) | 0.08 |
| | | (standard score) | 20.22 (1.52) | 20.43 (2.10) | 20.00 (0.00) | 0.22 |
| | | (age equivalence months) | 21.45 (3.98) | 22.27 (3.88) | 20.57 (3.95) | 0.06 |
| Preschool Language Scales - 5 | Auditory comprehension | (age equivalence months) | 20.84 (5.83) | 20.67 (6.24) | 21.03 (5.43) | 0.81 |
| | Expressive communication | (age equivalence months) | 18.29 (4.67) | 18.24 (4.65) | 18.35 (4.79) | 0.91 |

**Table 1.** *(Continued)*

| Measure | Variable | Value | DS (*n* = 79) Mean (SD) or *N* (%) | Control (*n* = 41) Mean (SD) or *N* (%) | JASPER-EMT (*n* = 38) Mean (SD) or *N* (%) | *P* value |
|---|---|---|---|---|---|---|
| Demographic (Caregiver) | Mother's age | (years) | 38.13 (6.15) | 37.95 (6.30) | 38.33 (6.06) | 0.79 |
| | Primary caregiver | Mother | 70 (91%) | 35 (90%) | 35 (92%) | 0.99 |
| | | Father | 7 (9%) | 4 (10%) | 3 (8%) | |
| | Primary language spoken at home | English | 60 (78%) | 31 (78%) | 29 (78%) | 0.36 |
| | | Spanish | 15 (19%) | 7 (18%) | 8 (22%) | |
| | | Other | 2 (3%) | 2 (5%) | 0 (0%) | |
| | Mothers Education | Less than 7th | 4 (5%) | 2 (5%) | 2 (5%) | 0.97 |
| | | Junior high | 6 (8%) | 2 (5%) | 4 (11%) | |
| | | Some high school | 3 (4%) | 2 (5%) | 1 (3%) | |
| | | High school | 7 (9%) | 3 (8%) | 4 (11%) | |
| | | Some college | 11 (14%) | 5 (12%) | 6 (16%) | |
| | | Special training | 2 (3%) | 1 (2%) | 1 (3%) | |
| | | College | 31 (40%) | 18 (45%) | 13 (35%) | |
| | | Graduate school | 13 (17%) | 7 (18%) | 6 (16%) | |
| | Father's education | Less than 7th | 5 (7%) | 4 (10%) | 1 (3%) | 0.06 |
| | | Junior high | 6 (8%) | 1 (3%) | 5 (14%) | |
| | | Some high school | 4 (5%) | 2 (5%) | 2 (6%) | |
| | | High school | 10 (13%) | 5 (13%) | 5 (14%) | |
| | | Some college | 15 (20%) | 11 (28%) | 4 (11%) | |
| | | Special training | 5 (7%) | 2 (5%) | 3 (8%) | |
| | | College | 17 (23%) | 11 (28%) | 6 (17%) | |
| | | Graduate school | 13 (17%) | 3 (8%) | 10 (28%) | |
| | Income | <$10,000 | 3 (4%) | 0 (0%) | 3 (9%) | 0.04 |
| | | $10,000–$19,999 | 7 (10%) | 6 (16%) | 1 (3%) | |
| | | $20,000–$29,999 | 7 (10%) | 3 (8%) | 4 (12%) | |
| | | $30,000–$39,999 | 8 (11%) | 3 (8%) | 5 (15%) | |
| | | $40,000–$49,999 | 6 (8%) | 4 (11%) | 2 (6%) | |
| | | $50,000–$59,999 | 5 (7%) | 3 (8%) | 2 (6%) | |
| | | $60,000–$79,999 | 6 (8%) | 2 (5%) | 4 (12%) | |
| | | $80,000–$100,000 | 7 (10%) | 1 (3%) | 6 (18%) | |
| | | >$100,000 | 23 (32%) | 16 (42%) | 7 (21%) | |

Comprehension age equivalence of 20.84 months ($SD$ = 5.83) and an Expressive Communication age equivalence of 18.29 months ($SD$ = 4.67), as measured by the PLS-5, and a MSEL Visual Receptive Age Equivalent of 22.51 months ($SD$ = 4.94) at baseline. Comparisons of key variables across sites revealed site differences for CA ($P < 0.0001$), PLS-5 Expressive Communication abilities ($P < 0.001$) and MSEL Visual Receptive Age Equivalent ($P < 0.0001$) at baseline, such that children at Site 2 were older and had lower language abilities than those at Site 1. Further details can be found in Table S1.

### *Children's BRIEF-P scores at baseline*

Means and standard deviations for BRIEF-P indices and subdomains across time and treatment groups are presented in Tables 2 and 3. On average, at baseline the sample had slightly elevated scores on FI and ISCI, with 22.9%% and 21.4% of participants receiving scores above the cutoff respectively. EMI

**Table 2** BRIEF-P *T*-scores per index across timepoints

| | Emergent Metacognition | | Flexibility Index | | Inhibitory Self Control | |
|---|---|---|---|---|---|---|
| | JASPER-EMT | Control | JASPER-EMT | Control | JASPER-EMT | Control |
| Entry | 70.11 (12.22) | 64.94 (11.51) | 57.03 (14.89) | 51.03 (10.86) | 59.14 (11.84) | 54.37 (11.00) |
| Exit | 65.21 (11.91) | 62.21 (13.21) | 53.25 (12.86) | 51.50 (9.61) | 56.21 (11.67) | 54.75 (13.41) |
| Follow-up | 68.36 (11.20) | 62.21 (13.21) | 51.70 (11.74) | 49.70 (11.86) | 55.64 (11.50) | 52.39 (10.68) |

*Note*: Exit timepoint is 3-month post-Entry. Follow-up timepoint is 6-month post-Entry.

scores were, on average, clinically elevated (with a T-score $> 65$); 60% of the participants had elevated EMI scores.

In comparing the subdomain scores (FI, ISCI and EMI) the ANOVA was significant ($F(2,207) = 23.43$, $P < 0.001$). The average EMI t-score was 13.50 units higher than the average FI t-score ($P < 0.001$) and 10.77 units higher than the average ISCI t-score ($P < 0.001$). The contrast between the FI and ISCI domains was not significant ($P = 0.39$) (Table 3).

*Treatment group differences.* The JASPER-EMT group had significantly higher scores on FI subdomain ($P = 0.05$) at baseline, differences between groups on EMI ($P = 0.07$) or ISCI ($P = 0.08$) approached significance. No group differences were found on the Emotion Control subdomain ($P = 0.31$), Planning and Organisation subdomain ($P = 0.19$) or Working Memory subdomain ($P = 0.13$) at entry. Group difference approached significance on the Inhibition subdomain ($P = 0.06$), and there were significantly higher scores in the Shifting subdomain at entry in the JASPER-EMT group compared with the Control group ($P = 0.04$).

*Site differences.* Comparisons of BRIEF-P scores across sites also revealed site differences found for scores on FI subdomain ($P < 0.001$) and ISCI subdomain ($P < 0.01$). Site differences were also detected for BRIEF-P indices, including the shifting ($P < 0.05$), emotion control ($P < 0.001$), working memory ($P = 0.05$) and inhibition ($P = 0.05$) indices. Site 1 was lower across each of these domains, likely explained by the relatively younger sample in Site 1 compared with Site 2. Further details can be found in Table S1.

## Predicting BRIEF-P scores at baseline

The overall regression equations explained a significant amount of variance in all three BRIEF-P indices (FI, $P < 0.001$; EMI, $P = 0.05$; ISCI, $P = 0.002$). The independent variables explained 39% of the variability in FI scores, 8% of the variability in EMI scores and 17% of the variability in ISCI scores. Within all three models (FI, EMI and ISCI), CA emerged as a significant predictor ($P < 0.01$, $P = 0.05$ and $P < 0.01$, respectively) of baseline scores. For ISCI scores at baseline, receptive language approached significance ($P = 0.06$) (Tables 4-6).

## Longitudinal models

Full model parameters are available in Tables 7–11.

### *Flexibility Index (FI)*

The 3-way interaction testing the effect of site on treatment over time was non-significant and was dropped from the model ($P > 0.05$). There was a significant time by treatment interaction from entry to exit on FI scores ($P = 0.01$), although these findings did not maintain to the 6-month follow-up. For children in the JASPER-EMT group, FI scores decreased on average 1.59 points between entry and exit but increased 3.43 points in the control group. On average, the JASPER-EMT group saw a reduction in FI scores from entry to follow up of 1.47 points while the control group saw an increase of 0.67 points (Figure 2).

*Shifting.* Further inspection of the subcomponents of the FI revealed that the time by treatment interaction was driven by a change in the shifting subdomain, which decreased by 2.2 points in the JASPER-EMT

**Table 3** BRIEF-P *T*-scores per subdomain across timepoints

| | Emotion Control | | Shifting | | Planning/Organisation | | Working Memory | | Inhibit | |
|---|---|---|---|---|---|---|---|---|---|---|
| | JASPER-EMT | Control | JASPER-EMT | Control | JASPER-EMT | Control | JASPER-EMT | Control | JASPER-EMT | Control |
| Entry | 52.06 (11.52) | 49.46 (10.03) | 57.86 (12.17) | 52.37 (10.69) | 63.89 (11.17) | 60.03 (13.10) | 70.97 (11.88) | 66.80 (10.96) | 62.54 (11.74) | 57.26 (12.02) |
| Exit | 49.65 (9.15) | 49.81 (13.37) | 53.68 (9.77) | 56.06 (12.39) | 62.38 (10.55) | 59.91 (11.67) | 65.15 (10.38) | 65.50 (11.14) | 59.18 (9.76) | 57.47 (12.45) |
| Follow-up | 48.85 (10.24) | 47.45 (10.24) | 54.64 (11.77) | 53.06 (13.04) | 63.88 (10.16) | 58.24 (12.50) | 69.70 (10.78) | 63.76 (12.88) | 59.70 (10.89) | 56.73 (12.51) |

*Note*: Exit timepoint is 3-month post-Entry. Follow-up timepoint is 6-month post-Entry.

**Table 4** Linear regression predicting *T*-scores on Flexibility Index at baseline

| | *B* | SE | *P* value |
|---|---|---|---|
| Intercept | 17.80 | 8.45 | 0.05 |
| Expressive language | 0.16 | 0.30 | 0.59 |
| Receptive language | −0.43 | 0.28 | 0.12 |
| Chronological age | 1.29 | 0.20 | <0.01 |
| Visual receptive | −0.27 | 0.32 | 0.40 |

**Table 5** Linear regression predicting *T*-scores on Emerging Metacognition Index at baseline

| | *B* | SE | *P* value |
|---|---|---|---|
| Intercept | 40.75 | 9.53 | <0.01 |
| Expressive language | 0.49 | 0.34 | 0.15 |
| Receptive language | −0.42 | 0.31 | 0.17 |
| Chronological age | 0.46 | 0.23 | 0.05 |
| Visual receptive | 0.36 | 0.37 | 0.33 |

**Table 6** Linear regression predicting *T*-scores on Inhibitory Self-Control Index at baseline

| | *B* | SE | *P* value |
|---|---|---|---|
| Intercept | 35.50 | 8.67 | <0.01 |
| Expressive language | 0.04 | 0.30 | 0.90 |
| Receptive language | −0.52 | 0.28 | 0.06 |
| Chronological age | 0.66 | 0.21 | <0.01 |
| Visual receptive | 0.30 | 0.33 | 0.38 |

group but increased by 3.3 points in the control group ($P = 0.03$). The three-way interaction between timepoint, treatment group and site for this model was non-significant and again dropped from the model ($P > 0.05$). The effect of time and time by treatment from entry to 6-month follow up was also not significant.

*Emotional control.* For the emotional control subdomain, the three-way interaction between timepoint, treatment group and site was

**Table 7** Longitudinal model predicting *T*-scores on Flexibility Index of BRIEF-P

| | *B* | SE | *P* value |
|---|---|---|---|
| Intercept | 22.18 | 6.57 | 0.001 |
| Site | 4.78 | 2.32 | 0.04 |
| Receptive language | −0.40 | 0.13 | 0.001 |
| Chronological age | 0.90 | 0.17 | <0.001 |
| Exit | 3.75 | 1.43 | 0.009 |
| Follow-up | 0.94 | 1.48 | 0.53 |
| Treatment | 6.62 | 2.41 | 0.007 |
| Time (exit)*Treatment | −4.85 | 1.96 | 0.01 |
| Time (follow-up)*Treatment | −2.19 | 1.98 | 0.27 |

*Note*: Site 1 is the reference group for Site, Entry is the reference group for Timepoint and the Control group is the reference group for Treatment. Exit timepoint is 3-month post-Entry. Follow-up timepoint is 6-month post-Entry.

**Table 8** Longitudinal model predicting *T*-scores on Emerging Metacognition Index of BRIEF-P

| | *B* | *SE* | *P* value |
|---|---|---|---|
| Intercept | 53.74 | 7.26 | <0.001 |
| Site | 4.51 | 2.55 | 0.08 |
| Receptive language | −0.24 | 0.15 | 0.10 |
| Chronological age | 0.32 | 0.18 | 0.09 |
| Exit | −0.05 | 1.73 | 0.98 |
| Follow-up | −1.56 | 1.78 | 0.38 |
| Treatment | 5.96 | 2.71 | 0.03 |
| Time (exit)*Treatment | −1.74 | 2.38 | 0.47 |
| Time (follow-up)*Treatment | 2.85 | 2.41 | 0.24 |

*Note*: Site 1 is the reference group for Site, Entry is the reference group for Timepoint, and the Control group is the reference group for Treatment. Exit timepoint is 3-month post-Entry. Follow-up timepoint is 6-month post-Entry.

non-significant and dropped from the model ($P > 0.05$). There was no effect of time or a time by treatment interaction from entry to exit, or from entry to 6-month follow up.

*Emergent Metacognition Index (EMI)*

The three-way interaction between timepoint, treatment group and site for the Emergent Metacognition Index was non-significant and so dropped from the model ($P > 0.05$). Both the time and time by treatment interaction terms were also non-significant from entry to exit and from exit to 6-month follow up indicating that EMI scores were both stable over time and the trajectories did not depend on treatment group.

**Table 9** Longitudinal model predicting *T*-scores on Inhibitory Self-Control Index of BRIEF-P

| | *B* | SE | *P* value |
|---|---|---|---|
| Intercept | 36.10 | 6.74 | <0.001 |
| Site | 8.17 | 3.45 | 0.02 |
| Receptive language | −0.30 | 0.13 | 0.03 |
| Chronological age | 0.53 | 0.17 | 0.002 |
| Exit | 0.84 | 2.13 | 0.69 |
| Follow-up | 1.34 | 2.13 | 0.53 |
| Treatment | 8.17 | 3.45 | 0.007 |
| Time (exit)*Treatment | −3.73 | 2.89 | 0.20 |
| Time (follow-up)*Treatment | −4.30 | 2.86 | 0.14 |
| Time (exit)*Site | 4.99 | 3.06 | 0.11 |
| Time (follow-up)*Site | −2.37 | 3.03 | 0.44 |
| Time (exit)*Treatment*Site | 2.23 | 4.37 | 0.61 |
| Time (follow-up)*Treatment*Site | 10.14 | 4.48 | 0.02 |

*Note*: Site 1 is the reference group for Site, Entry is the reference group for Timepoint and the Control group is the reference group for Treatment. Exit timepoint is 3-month post-Entry. Follow-up timepoint is 6-month post-Entry.

**Table 10** Longitudinal model predicting *T*-scores on attentional shifting subdomain of Flexibility Index of BRIEF-P

| | *B* | SE | *P* value |
|---|---|---|---|
| Intercept | 40.85 | 9.26 | <0.001 |
| Site | 1.96 | 2.72 | 0.47 |
| Receptive language | −0.36 | 0.22 | 0.10 |
| Chronological age | 0.52 | 0.21 | 0.01 |
| Exit | 3.03 | 1.74 | 0.08 |
| Follow-up | 0.23 | 1.72 | 0.89 |
| Treatment | 4.03 | 2.80 | 0.15 |
| Time (exit)*Treatment | −5.42 | 2.46 | 0.03 |
| Time (follow-up)*Treatment | −2.54 | 2.45 | 0.30 |

*Note*: Site 1 is the reference group for Site, Entry is the reference group for Timepoint and the Control group is the reference group for Treatment. Exit timepoint is 3-month post-Entry. Follow-up timepoint is 6-month post-Entry.

**Table 11** Longitudinal model predicting *T*-scores on emotional control subscale of Flexibility Index of BRIEF-P

| | *B* | SE | *P* value |
|---|---|---|---|
| Intercept | 29.63 | 7.76 | <0.01 |
| Site | 4.49 | 2.28 | 0.05 |
| Receptive language | −0.29 | 0.18 | 0.11 |
| Chronological age | 0.74 | 0.17 | <0.001 |
| Exit | −0.12 | 1.45 | 0.91 |
| Follow-up | −1.87 | 1.44 | 0.20 |
| Treatment | 2.53 | 2.35 | 0.29 |
| Time (exit)*Treatment | −1.39 | 2.06 | 0.50 |
| Time (follow-up)*Treatment | −0.09 | 2.05 | 0.97 |

*Note*: Site 1 is the reference group for Site, Entry is the reference group for Timepoint and the Control group is the reference group for Treatment. Exit timepoint is 3-month post-Entry. Follow-up timepoint is 6-month post-Entry.

*Inhibitory Self Control Index (ISCI)*

There was a significant three-way interaction between time, treatment and site in the model predicting ISCI, driven by a significantly larger decrease in ISCI scores between exit and follow-up for children in the control group of Site 2 compared with Site 1 (see Figure 3). There was an estimated 6.87 point decrease in ISCI scores in the control group of Site 2 compared with a 0.49 point increase in the control group of Site 1. All other interactions were found to be nonsignificant for the control group. No significant effect of site on treatment was found for children in the JASPER-EMT group at any timepoint ($P > 0.05$). For children in the JASPER-EMT group, time and time by treatment interaction terms were non-significant from entry to exit and from exit to

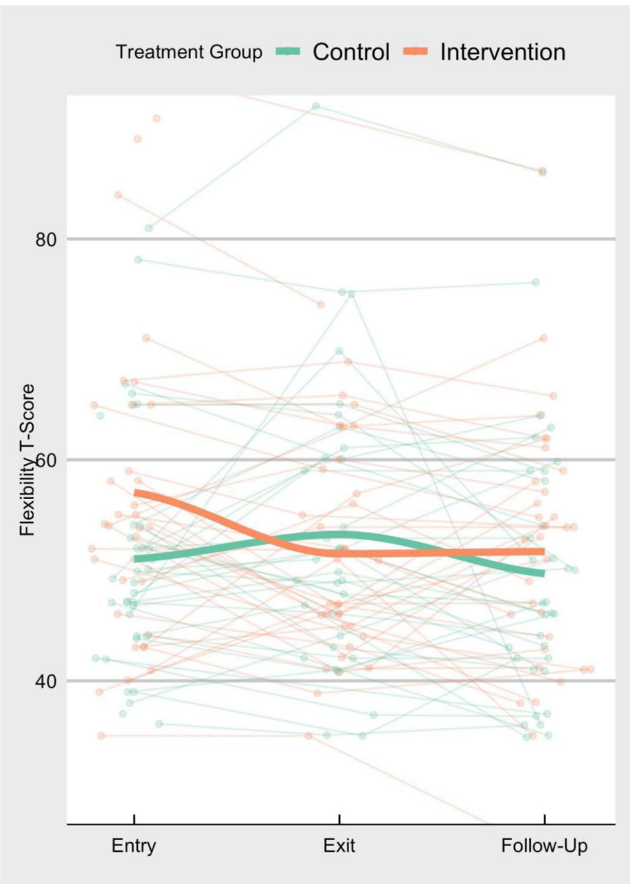


**Figure 2.** Change in Flexibility Index T-Scores across timepoints. Exit timepoint is 3-month post-Entry. Follow-up timepoint is 6-month post Entry.

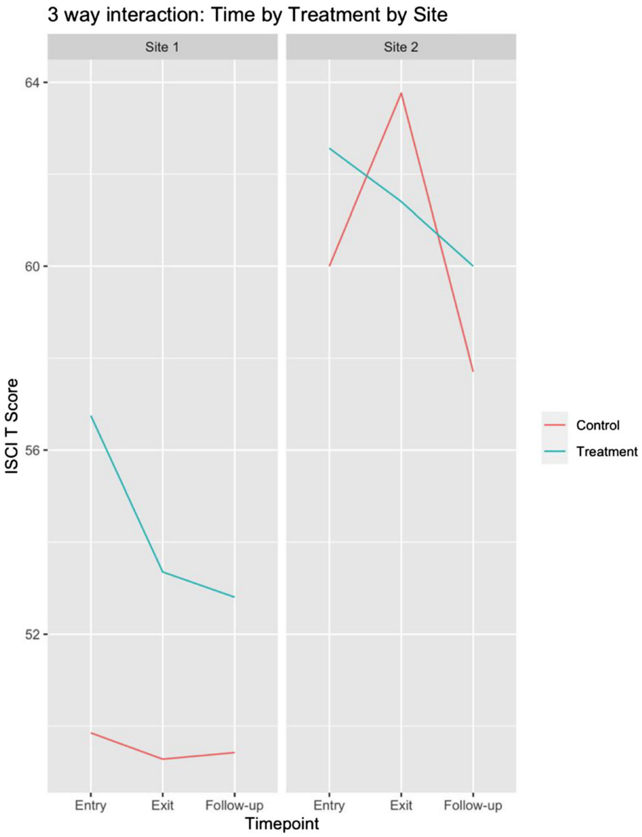


**Figure 3.** Three-way interaction between Site, Treatment Group and Time for *T*-scores on ISCI. Exit timepoint is 3-month post Entry. Follow-up timepoint is 6-month post Entry.

6-month follow up for children's ISCI scores, indicating that these scores were stable over time.

## Discussion

The present study characterised EF abilities among a sample of preschool-aged children with DS and explored the effect of a naturalistic early social communication intervention (JASPER-EMT) on EF abilities over time. Findings indicate that evidence-based social communication interventions (e.g. JASPER-EMT) may have a short-term impact on parent-reported EF skills among young children with DS, namely, in the areas of flexibility and attentional shifting. Attentional shifting is an EF skill that appears to worsen with age among individuals with DS (Loveall *et al.* 2017), so it is promising that intervention may successfully support these skills. Additionally, our results suggest that preschoolers with DS in this sample have relative weaknesses in emergent metacognition, driven by challenges in working memory. Most EF skills remained relatively stable over time for both treatment groups, with two exceptions detailed in the sections that follow.

### Baseline characteristics of preschoolers with DS

At baseline, children in this sample had an average chronological age (CA) of 39 months, with children at Site 1 slightly younger (36 months) than those at Site 2 (43 months). In addition to differences in CA, children at Site 1 had higher age equivalent scores on the Expressive Communication and Auditory Comprehension subdomains of the PLS-5 and Visual Receptive age equivalent scores on the MSEL Visual Receptive Domain. Overall, children at Site 2 were older and had lower language abilities than their counterparts at Site 1, although it must be noted that children at both sites exhibited significant language delays. Differences also appeared in the number of bilingual children enrolled at each site, with 82.5% of children at Site 2 identified as having more than one language spoken in the home, in contrast with 22% of those at Site 1. Given that bilingualism was determined by number of languages spoken in the home, rather than language spoken by the child, as well as disparities between sites in the number of families who identified more than one language being spoken in the home, it is difficult to ascertain the impact of bilingualism on the development of EF in this sample. Given that bilingualism has been found to have an impact on the development of EF among TD children (e.g. Foy & Mann 2013; Crivello *et al.* 2016; Tran *et al.* 2019), it is important to further examine the impact of bilingualism on the development of EF in children with DS. This is an important next step for future work on EF in samples of children with DS and other neurodevelopmental disabilities.

Children's CA at baseline predicted EF skills across all three indices of the BRIEF-P, consistent with age effects, which have been established in samples of older children with DS (Loveall *et al.* 2017). Children's expressive language and visual receptive abilities did not predict baseline scores. The lack of a significant relationship between children's expressive language abilities and their EF skills may be explained by the low level, and lack of variability, of language exhibited by participants in this sample. The finding that CA was a significant predictor of children's EF abilities across all indices of the BRIEF-P may indicate that the age range at entry may have played a role in children's baseline EF abilities, underscoring the role played by children's experience over time on EF abilities.

## Baseline profiles of EF

Comparisons of children's scores on the BRIEF-P indices revealed significant differences, such that children's scores on EMI were significantly higher (more impacted) than scores on both FI and ISCI. This finding is consistent with previous studies using samples of young children with DS (Lee *et al.* 2011; Daunhauer *et al.* 2014; Loveall *et al.* 2017) that suggests that preschool aged children with DS may present uneven profiles of EF. Clinically significant challenges in working memory were observed, as well as relative strengths in attentional shifting. These findings are consistent with findings reported by Loveall *et al.* (2017), in which young children with DS (aged 2–5 years) were found to have relative strengths in emotional control and attentional shifting and relative weaknesses in working memory.

## Changes in EF profiles over time

The analysis of children's EF profiles across three timepoints revealed that children in the waitlist control group exhibited relatively stable profiles over the course of the 6-month study. For scores on EMI and FI, children in the control group exhibited little to no change over the course of the study, except for the finding that ISCI improved between exit and follow-up. Between exit and the 6-month follow up, children in the control group at Site 2 exhibited a significant decrease (indicating improvement) in ISCI scores, although the same was not seen for Site 1. Despite sampling criteria aimed at recruiting a developmentally homogeneous sample, site differences emerged across many key variables, including CA, expressive language abilities and visual receptive age equivalent. As such children at Site 2 were older and more developmentally delayed than children at Site 1 and were more likely to be enrolled in preschool and to be receiving behavioural intervention. It is possible that attending preschool and receiving behavioural intervention may have contributed to changes in children's scores on ISCI, especially because this index measures children's ability to control their emotions and behaviours, both areas that could be impacted by schooling and behavioural intervention. Although this is one possible explanation for the significant three-way interaction, it is difficult to ascertain the specific cause for the difference seen in children in the control group at Site 2.

Children who received JASPER-EMT exhibited significant decreases (indicating improvements in performance) in FI scores between entry and exit, although no significant changes were seen for either EMI or ISCI scores. These improvements in FI correspond with improvements in expressive and receptive language abilities for children receiving JASPER-EMT (Kaiser 2017). More specifically, children in the treatment group exhibited significant changes in scores on the PLS-5 Auditory Comprehension and Expressive Communication domains from entry to exit, as well as an increase in use of spontaneous language (i.e. commenting). The concurrent nature of the improvements in expressive and receptive language abilities and children's FI scores suggests that improvements in children's language may have contributed to the improvements seen in their performance on FI, although further research is needed to explore this relationship. It must be noted that the treatment effect for FI did not maintain at the 6-month follow up, although there were improvements over time. This drop-off in scores on more distal intervention outcomes from exit to follow-up time points is a phenomenon common to intervention studies (e.g. Landa *et al.* 2011; Hampton *et al.* 2020; Kasari *et al.*, 2015). Overall, the combination of improvements in both language and children's scores on FI during the active treatment period, as well as the lack of improvements on the other indices for children receiving JASPER-EMT, support the potential impact of JASPER-EMT on children's EF abilities, specifically with regard to flexibility.

A closer inspection of the subdomains of FI revealed that improvements in FI scores for children in the JASPER-EMT group were driven by improvements in attentional shifting. This finding is of note given evidence to suggest that attentional shifting may become more challenging with age among children with DS. In a recent cross-sectional study of 2- to 18-year-old children with DS, researchers found that the development of attentional shifting followed a unique developmental trajectory (Loveall *et al.* 2017). Whereas attentional shifting was found to be a relative strength among children 2–5 years of age, the opposite was observed among children aged 6 to 18 years of age, suggesting a possible delay in the development of attentional shifting in this population. For children who received JASPER-EMT, improvements seen in attentional shifting may be reflective of increases in their ability to engage with social partners during a shared activity, a skill explicitly targeted within the JASPER-EMT intervention. Additionally, increased awareness of one's social partner and engagement in reciprocal interactions may bolster the child's ability to focus their attention on the task at hand. Lastly, behavioural strategies implemented as a part of the JASPER-EMT approach could also contribute to the children's increased attention to relevant cues through increasing children's understanding of behavioural contingencies.

Findings of the current study suggest that attentional shifting in young children with DS, as reported by parents, may be a malleable skill in children with DS. Further research is needed to understand the mechanism through which change is affected in attentional shifting abilities, and whether these changes can be supported after the active treatment period.

## Limitations

While this study extends the current literature by replicating EF profiles in the largest sample of young children with DS to date, as well as examining the effects of a naturalistic behavioural intervention on children's EF abilities over time, some limitations must be acknowledged. First, this study did not include a comparison group of children with either typical development or with other neurodevelopmental disorders. Therefore, it cannot be determined whether the EF profiles and trajectories found in this study are unique to the Down syndrome population. Further, no cross-informant, observational or task-based ratings of EF were collected. Because the current study relied solely on the BRIEF-P as a measure of EF, we must also consider the possibility that parents' reports of their children's EF abilities may be coloured by extraneous factors, including knowledge of children's intervention group assignment. It is possible, for instance, that parents whose children received intervention were more likely to report improvements in their children's EF abilities, specifically on the flexibility Index, than parents whose children were assigned to the waitlist control group. Using multiple measures would increase confidence that the observed patterns are not driven by method of measurement or informant. Lastly, significant differences in individual characteristics emerged between sites, which possibly contributed post-intervention changes in ISCI scores for children in the control group at Site 2. This finding raises questions regarding the impact of individual differences on the development of EF over time, as well as how the individual differences may impact the effect of intervention. Additionally, groups were significantly different on the Flexibility Index at baseline and approached significance on the EMI and ISCI at baseline. The time by treatment effect on the

Flexibility Index and absence of a significant effect on the other two indices, combined with findings of concurrent improvements in language outcomes, strengthens our confidence that baseline group differences alone did not drive the significant interaction effect.

## Conclusions

Little is known regarding EF abilities in very young children with DS, because the majority of studies exploring EF in individuals with DS have focused on older children, adolescents and adults. This study expands upon the literature by exploring EF profiles in a sample of very young children with DS and examines the impact of a naturalistic social communication intervention (JASPER-EMT) on children's EF abilities over time. Children exhibited differential abilities across the three indices of the BRIEF-P at baseline, with relative strengths on FI and ISCI, and relative weaknesses on EMI. This study also offers preliminary findings regarding the impact of an existing early social communication intervention on EF abilities among young children with DS. Given the importance of EF on all domains of children's development and that EF skills are largely impacted in adults with DS, these findings merit further exploration and raise the possibility of leveraging evidence-based language interventions to facilitate the development of EF skills in children with DS.

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## Conflict of interest

The authors declare no conflicts of interest.

## Ethics approval statement

The Institutional Review Boards (IRB) at the University of California, Los Angeles and Vanderbilt University approved the current study. All participants in the study provided parental consent.

## Data availability statement

The datasets generated and/or analysed during the current study are not publicly available due to privacy concerns for minors with disabilities.

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## Supporting Information

Additional Supporting Information may be found online in the supporting information tab for this article.

**Table S1:** Descriptive Statistics and Comparisons of Participant Characteristics at Baseline.
**Figure S1:** Change in Inhibitory Self Control Index T-Scores across timepoints.
**Figure S2:** Change in Emerging Metacognition Index T-Scores across timepoints.
**Figure S3:** Change in Shifting Subdomain T-Scores across timepoints.
**Figure S4:** Change in Emotion Control Subdomain T-Scores across timepoints.
**Figure S5:** Change in Inhibition Subdomain T-Scores across timepoints.
**Figure S6:** Change in Working Memory Subdomain T-Scores across timepoints.
**Figure S7:** Change in Planning and Organisation Subdomain T-Scores across timepoints.