

Special Issue

Language and executive functioning in young adults with Down syndrome

K. Kristensen,¹ K. M. Lorenz,¹ X. Zhou,¹ B. Piro-Gambetti,^{1,2} S. L. Hartley,^{1,2}  S. P. Godar,¹ S. Diel,¹ E. Neubauer¹ & R. Y. Litovsky¹

¹ Waisman Center, University of Wisconsin–Madison, Madison, WI, USA

² School of Human Ecology, University of Wisconsin–Madison, Madison, WI, USA

Abstract

Background This study examined the association between executive functioning and language in young adults with Down syndrome (DS).

Method Nineteen young adults with DS (aged 19–24 years) completed standardised measures of overall cognition, vocabulary, verbal fluency and executive function skills.

Results Friedman's analysis of variance ($\chi^2(3) = 28.15, P < .001$) and *post hoc* comparisons indicated that, on average, participants had a significantly lower overall non-verbal than verbal cognitive age equivalent and lower expressive than receptive vocabulary skills. Using Spearman correlations, performance on a verbal measure of cognition inhibition was significantly negatively related to receptive vocabulary ($\rho = -.529$, adjusted $P = .036$) and verbal fluency ($\rho = -.608$, adjusted $P = .022$). Attention was significantly positively correlated with receptive ($\rho = .698$, adjusted- $p = .005$) and expressive ($\rho = .542$, adjusted $P = .027$) vocabulary. Verbal working memory was significantly positively associated with receptive vocabulary

($\rho = .585$, adjusted $P = .022$) and verbal fluency ($\rho = .737$, adjusted $P = .003$). Finally, visuospatial working memory was significantly associated with receptive vocabulary ($\rho = .562$, adjusted $P = .027$).

Conclusions Verbal and non-verbal measures of executive functioning skills had important associations with language ability in young adults with DS. Future translational research is needed to investigate causal pathways underlying these relationships. Research should explore if interventions aimed at increasing executive functioning skills (e.g. attention, inhibition and working memory) have the potential to lead to increases in language for young adults with DS.

Keywords Down syndrome, executive functioning, language, memory, vocabulary

Introduction

Down syndrome (DS) is caused by a full or partial third copy of chromosome 21 and is one of the most common genetic disorders associated with intellectual disability (ID), occurring in about 1 in 700 live births each year in the United States (Martin *et al.*, 2009). Children with DS experience challenges with language beyond what would be expected given their

Correspondence: Ms Kayla Kristensen, Waisman Center, University of Wisconsin–Madison, 1500 Highland Ave, Madison, WI 53705, USA. (e-mail: kayla.kristensen@wisc.edu)

level of ID (Grieco *et al.*, 2015; Naess *et al.*, 2011; Wittey & Penke, 2017). Although less studied, these relative deficits in language have been reported to continue into adulthood in DS (Grieco *et al.*, 2015; Wittey & Penke, 2017). Language has marked consequences for adult quality of life, as it influences opportunities and experiences in employment, independent living and community involvement, as well as family and social relationships. Efforts to identify mechanisms associated with language in DS are thus critical for enhancing adult quality of life in DS.

A substantial body of research has examined the language functioning of children with DS. Delays in language begin early in life and include a slower transition from babbling to spoken words, reduced expressive vocabulary and difficulties with understanding and use of grammatical markers (Chapman & Hesketh, 2000; Roberts *et al.*, 2007). Relative to same-aged peers without DS, school-aged children with DS demonstrate challenges in processing, understanding and producing language (Grieco *et al.*, 2015; Lukowski *et al.*, 2019). In many of these studies, impairments in expressive language in children with DS were greater than those in receptive language, with the former below what would be expected given non-verbal mental age (Abbeduto *et al.*, 2003; Naess *et al.*, 2011). There is substantially less research describing the language ability of young adults with DS. The few studies on this topic report that expressive language may continue to be below receptive language level and below what would be expected given non-verbal mental age (Martin *et al.*, 2009; Naess *et al.*, 2011). A better understanding of language strengths and areas of need in DS is necessary to inform language supports and interventions. In addition to impairments in language, children with DS demonstrate difficulties with executive functioning (Carney *et al.*, 2013; Lanfranchi *et al.*, 2010; Rowe *et al.*, 2006). Executive functioning involves cognitive processes used in goal-driven behaviour and includes working memory (temporary storing and manipulating information), focus (directing and shifting attention), cognitive flexibility (switching between tasks) and inhibition (overriding dominant or automatic responses) (Dawson & Guare, 2004; Miyake *et al.*, 2000). In typically developing populations, executive functioning generally matures between early adolescence and young adulthood (Hoyo *et al.*, 2015;

O'Hare & Sowell, 2008), in keeping with the developing prefrontal cortex (Lynch *et al.*, 2019). Relative to typically developing peers of chronological and non-verbal mental age, children with DS demonstrate poor verbal working memory (Carney *et al.*, 2013; Godfrey & Lee, 2018; Lanfranchi *et al.*, 2010), as well as deficits in inhibition (Borella *et al.*, 2013; Pennington *et al.*, 2003). Much less is known about the executive functioning of young adults with DS prior to ageing and dementia-related declines in middle and older adulthood. Rowe *et al.* (2006) described difficulties with verbal and visual short-term memory for adults with DS. Traverso *et al.* (2018) found that in a sample of children and adults with DS ($M = 14$ years, range = 6–24 years), inhibition skills were markedly below that of typically developing peers. Moreover, compared with typically developing peers of the same mental age, children, adolescents and young adults with DS demonstrated difficulties with verbal working memory (Costanzo *et al.*, 2013; Seung & Chapman, 2000).

Theoretical and empirical evidence from populations without DS suggests that language ability has important connections with executive functioning (e.g. Baddeley, 2012; Ellis Weismer *et al.*, 2005; Vugs *et al.*, 2014). Much of the evidence for the association between executive functioning and language comes from studies on typically developing populations (e.g. Woodard *et al.*, 2016). For example, inhibition is associated with better lexical and syntactic ability in children and young adults (Khanna & Boland, 2010), whereas working memory positively predicts auditory sentence comprehension in children (Roberts *et al.*, 2007) and sentence production ability in young adults (Slevc, 2011). Evidence on the association between executive functioning and language is also found in populations with specific language impairments. For example, children with specific language impairment have difficulties with inhibition (Bishop & Norbury, 2005), task shifting (Marton, 2008) and working memory (Ellis Weismer *et al.*, 2005; Vugs *et al.*, 2014). Most of the above studies assessed executive functioning using verbal tasks (i.e. tasks that relied on verbal instructions and responses), raising concern that reported associations between executive functioning and language could be conflated due to shared reliance on verbal language ability. However, non-verbal tasks of working

memory, focus, cognitive flexibility and inhibition have also been found to be associated with receptive and expressive language (Kaushanskaya *et al.*, 2017).

The extent to which executive functioning is associated with language in young adults with DS is largely unknown as only a handful of studies have examined this association. In a study of young and middle-aged adults with DS, better verbal working memory was associated with better semantic and phonemic verbal fluency (Stavroussi *et al.*, 2016). In a longitudinal study, Laws and Gunn (2004) found that verbal short-term memory, assessed through digit span and non-word repetition tasks, was a strong positive predictor of receptive vocabulary and grammar comprehension in young adults with DS. Furthermore, Faught and Connors (2019) found an association between verbal tasks of executive functioning (sustained attention, inhibition and short-term memory) and vocabulary and syntax in individuals with DS. Understanding the associations between verbal and non-verbal components of executive functioning and language ability can inform formal (e.g. therapies) and informal (e.g. caregiver efforts) for enhancing language in young adults with DS.

The current study had three aims related to young adults with DS: (1) Describe language ability on measures of receptive and expressive vocabulary and verbal fluency; (2) evaluate the relationship between these language measures and overall cognitive mental age; and (3) determine the association between language and verbal and non-verbal executive functioning. We hypothesised that young adults with DS would exhibit receptive vocabulary skills comparable with that of their non-verbal mental age. In line with studies on children with DS (Abbeduto *et al.*, 2003; Naess *et al.*, 2011), we hypothesised that expressive language ability would be below that of non-verbal mental age. Lastly, we hypothesised that there would be a positive association between performance on verbal and non-verbal measures of executive functioning and language.

Method

Participants

Nineteen young adults with DS aged 19–24 years (10 female and 9 male) participated in the study.

Participants were recruited as part of a larger study on hearing and cognition in DS. Recruitment occurred through study flyers shared with state and local intellectual and developmental disability centres, as well as a regional DS research registry. Inclusion criteria included English as a primary language and use of spoken two- to three-word phrases as the larger planned study required multi-word responses. Exclusion criteria included the presence of a medical condition that impacted cognitive testing (e.g. use of noisy oxygen device that interfered with hearing testing) or cognitive ability (e.g. untreated cardiovascular conditions). Informed consent was completed by the participant and/or their legal guardian. Table 1 provides socio-demographic information.

Study procedure

Participants and a caregiver attended two or more study visits across multiple days to complete testing (approximately 12 h). Participants were reimbursed for travel expenses and received \$140 for the larger study. Caregivers completed questionnaires about the developmental history of the young adult with DS, and participants completed directly administered assessments of overall cognitive ability, receptive and expressive vocabulary, verbal fluency and executive functioning. The study was approved by the University of Wisconsin - Madison Institutional Review Board.

Measures

Overall cognitive ability

The Kaufman Brief Intelligence Test Second Edition (KBIT-2; Kaufman & Kaufman, 2004) is a measure of verbal and non-verbal intelligence and is normed on individuals aged 4–90 years. This measure has been used with adults with DS (e.g. Hamburg *et al.*, 2019). The verbal and non-verbal intelligence standard scores ($M = 100$, $SD = 15$) and mental age equivalent (in years) were analysed.

Receptive language

The Peabody Picture Vocabulary Test Fifth Edition (PPVT-5; Dunn, 2019) measures receptive vocabulary in individuals 2.5–90+ years. This measure is reliable and valid for individuals with ID

Table 1 Participant socio-demographic information

Type of DS (n, %)	
Trisomy 21	15 (79)
Mosaicism	1 (5)
Translocation/partial	1 (5)
Unknown	2 (11)
Chronological age in years (M, SD; range)	22.20, 1.70; 19–24
Biological sex	
Male (n, %)	9 (47)
Ethnicity	
Non-Hispanic (n, %)	18 (95)
Hispanic (n, %)	0 (0)
Unknown (n, %)	1 (5)
Race	
Caucasian (n, %)	15 (79)
African American (n, %)	1 (5)
More than 1 race (n, %)	2 (11)
Unknown (n, %)	1 (5)
Hearing status ^a (n, %)	
Normal	9 (47)
Mild-to-moderate hearing loss	9 (47)
Unilateral profound hearing loss	1 (5)
KBIT-2	
Non-verbal mental age in years (M ± SD; range)	5.58 ± 2.41; 4.00–11.67
Verbal mental age in years (M ± SD range)	6.88 ± 2.70; 4.00–11.33
PPVT-5	
Receptive vocabulary age in years (M ± SD; range)	8.13 ± 2.89; 3.17–15.33
EVT-3	
Expressive vocabulary age in years (M ± SD; range)	7.04 ± 2.74; 3.00–15.50

EVT-3, Expressive Vocabulary Test Third Edition (Williams, 2019); KBIT-2, Kaufman Brief Intelligence Test Second Edition (Kaufman & Kaufman, 2004); PPVT-5, Peabody Picture Vocabulary Test Fifth Edition (Dunn, 2019).

^aHearing status was examined using a pure-tone hearing threshold test through air conduction and bone conduction as appropriate. Two individuals with moderate hearing loss used hearing amplification. The individual with unilateral profound hearing loss did not use amplification and had normal to borderline normal hearing in the unaffected ear.

(Esbensen *et al.*, 2017). Individuals point to pictures that illustrate words. The standard scores ($M = 100$, $SD = 15$) and vocabulary age equivalent (in years) were analysed.

Expressive language

The Expressive Vocabulary Test Third Edition (EVT-3; Williams, 2019) is a measure of expressive vocabulary and word retrieval for individuals 2.5–

90+ years. The EVT-3 is a reliable and valid tool for individuals with ID (Esbensen *et al.*, 2017).

Participants say a word to label a picture. The standard scores ($M = 100$, $SD = 15$) and vocabulary age equivalent (in years) were analysed.

Verbal fluency

The Developmental Neuropsychological Testing Second Edition, Word Generation Semantic Fluency Subtest (NEPSY-2; Korkman *et al.*, 2007) is a measure of verbal fluency. This task is appropriate for adults with DS (Esbensen *et al.*, 2017). Participants name as many different animals as possible in 1 min. Total number of animals named was analysed.

Executive functioning

Four tests were used to measure executive functioning. First, the Cat and Dog Stroop task (Ball *et al.*, 2008) is an adapted version of a Stroop test assessing cognitive inhibition, which is appropriate for adults with DS (Esbensen *et al.*, 2017) and has previously correlated with other measures of executive functioning in adults with DS (Ball *et al.*, 2008; Hartley *et al.*, 2017). Participants point to and name pictures of cats and dogs as quickly as possible. Naming time is recorded. Next, participants switch the labels (e.g. call the cat 'dog') as quickly as possible. Switching time and number of errors are recorded. The switching trial time was analysed. Second, the Wechsler Intelligence Scale for Children Fourth Edition – Digit Span Forward (WISC-IV; Wechsler, 2003) assesses attention and has been found to have adequate test properties in young and middle-aged adults with DS (Hartley *et al.*, 2017; Seung & Chapman, 2000). Individuals repeat a series of digits. The WISC-IV Total Score was analysed. Third, the WISC-IV Digit Span Backward is a measure of verbal working memory used with middle-aged adults with DS (Hartley *et al.*, 2017). Participants remember and then repeat a series of digits in reverse order. The WISC-IV Total Score was used analysed. Fourth, the Corsi Block Tapping Task – Backward (Wechsler *et al.*, 2004) is a measure of visuospatial short-term working memory that has been used with young and middle-aged adults with DS (Hartley *et al.*, 2017; Yang *et al.*, 2014). Participants first watch an

examiner tap a series of blocks and are then asked to tap those same blocks in reverse order. The total number correct was analysed.

Data analysis plan

Shapiro–Wilk tests were performed to examine the distribution of study variables. Results from some assessments were not normally distributed, including KBIT-2 non-verbal mental age ($W = 0.770$, $P < .001$), EVT-3 mental age ($W = 0.763$, $P < .001$), EVT-3 standard scores ($W = 0.871$, $P = .015$) and WISC-IV Digit Span Forward ($W = 0.793$, $P = .001$) and Backward scores ($W = 0.877$, $P = .023$). Hence, non-parametric analyses were conducted. A Friedman's analysis of variance was conducted to examine the differences between four age equivalent measures. Bonferroni correction was conducted to adjust P -values for *post hoc* multiple comparisons. To examine whether there were deficits in receptive or expressive vocabulary relative to overall cognition, aligned ranks transformation of analysis of variance (art ANOVA) was conducted for KBIT-2, PPVT-5 and EVT-3 mental age equivalency scores. Spearman correlations were then conducted to examine the association between the KBIT-2 verbal and non-verbal mental age scores and language ability (PPVT-5, EVT-3/NEPSY-2). Finally, Spearman correlations were conducted between language ability (PPVT-5, EVT-3 and NEPSY-2) and measures of executive functioning (Cat and Dog Stroop, WISC-IV Digit Span Forward and Backward and WISC-IV Integrated Corsi Block Tapping Task – Backward). Benjamini–Hochberg correction was performed to adjust P -values for multiple correlations ($n = 18$).

Results

Missing data were minimal. One participant was unable to understand instructions for the switch trial of the Cat and Dog Stroop task. This individual was excluded from analysis of Cat and Dog Stroop task but included in analyses of other measures. The following floor effects (i.e. lowest possible score) were observed: Cat and Dog Stroop task ($n = 1$), EVT-3 ($n = 1$), WISC-IV Digit Span Forward ($n = 1$), WISC-IV Digit Span Backward ($n = 6$), Corsi Block Tapping Task – Backward ($n = 2$), NEPSY-2 ($n = 2$),

PPVT-5 ($n = 3$), KBIT-2 Non-verbal ($n = 7$) and KBIT-2 Verbal ($n = 6$). Table 2 provides the sample mean and SD, as well as the range of average normed scores for the typically developing population when available. On average, participants' standard scores on the PPVT-5 and EVT-3 equated to age equivalencies of 8.13 years (ranging 3.17–15.33) and 7.04 years (ranging 3.00–15.50) on the PPVT-5 and EVT-3, respectively.

Figure 1 plots the KBIT-2, EVT-3 and PPVT-5 age equivalents. The mean (markers) and SD (bars) for each age equivalent score across participants are shown on the rightmost portion of the plot. Results from a Friedman test found a significant effect of age equivalency by test ($\chi^2(3) = 28.15$, $P < .001$). *Post hoc* analysis with Bonferroni correction revealed significantly lower KBIT-2 non-verbal mental age than KBIT-2 verbal mental age ($P = .048$), EVT-3 expressive vocabulary age ($P = .048$) and PPVT-5 receptive vocabulary age ($P < .001$). Results also identified significantly lower EVT-3 and KBIT-2 verbal ages than PPVT-5 age (both with $P = .048$).

As shown in Figure 2, there was a significant positive correlation between the KBIT-2 verbal mental age and the PPVT-5 ($\rho = .821$, adjusted $P < .001$), EVT-3 ($\rho = .576$, adjusted $P = .022$) and NEPSY-2 ($\rho = .636$, adjusted $P = .014$). There was a significant positive association between KBIT-2 non-verbal mental age and the PPVT-5 ($\rho = .629$, adjusted $P = .014$) and EVT-3 ($\rho = .564$, adjusted $P = .024$) but not with NEPSY-2 ($\rho = .273$, adjusted $P = .290$).

Spearman correlations were used to assess the association between executive functioning and language. As shown in Figure 3, the Cat and Dog Stroop switching time was significantly negatively associated with the PPVT-5 ($\rho = -.529$, adjusted $P = .036$) and NEPSY-2 ($\rho = -.608$, adjusted $P = .022$) but not with EVT-3 ($\rho = -.224$, adjusted $P = .370$).

Figure 4 shows the associations between the three other measures of executive functioning and language. WISC-IV Digit Span Forward was significantly positively correlated with the PPVT-5 ($\rho = .698$, adjusted $P = .005$) and the EVT-3 ($\rho = .542$, adjusted $P = .027$) but not with the NEPSY-2 ($\rho = .247$, adjusted $P = .330$). The WISC-IV Digit Span Backward was significantly positively correlated with the PPVT-5 ($\rho = .585$,

Table 2 Descriptive statistics for measures of language ability and executive functioning

Ability	Measure	Type of score	Young adults with DS (mean ± SD)	Normal score range
Verbal and non-verbal intelligence	KBIT-2	Verbal SS	51.44 ± 12.94	85–115
		Non-verbal SS	52.11 ± 14.96	85–115
		IQ composite SS	48.22 ± 12.01	90–109
Receptive language	PPVT-5	SS	58.36 ± 14.52	85–115
Expressive language	EVT-3	SS	61.22 ± 12.44	85–115
Verbal fluency	NEPSY-2 animal category	Raw score (TS)	8.61 ± 5.53	NA
Attention	WISC-IV Digit Span Forward	Forward TS	3.26 ± 1.44	NA
		Forward longest span	2.78 ± 0.97	NA
		Backward TS	2.47 ± 2.41	NA
Verbal short-term working memory	WISC-IV Digit Span Backward	Backward longest span	1.73 ± 1.32	NA
		Backward longest span	2.65 ± 1.87	NA
Cognitive inhibition	Cat and Dog Stroop	Switching time (seconds)	26.93 ± 15.21	NA
		Naming errors	0.22 ± 0.73	NA
		Switching errors	0.50 ± 0.99	NA
		Backward TS	2.65 ± 1.87	NA
Visuospatial short-term working memory	WISC-IV Integrated Corsi Block Tapping Task – Backward	Backward longest span	2.65 ± 1.06	NA

EVT-3, Expressive Vocabulary Test Third Edition (Williams, 2019); KBIT-2, Kaufman Brief Intelligence Test Second Edition (Kaufman & Kaufman, 2004); NA, not applicable; NEPSY-2, Developmental Neuropsychological Testing Second Edition, Word Generation Semantic Fluency Subtest (Korkman *et al.*, 2007); PPVT-5, Peabody Picture Vocabulary Test Fifth Edition (Dunn, 2019); SS, standard score; TS, total score; WISC-IV Integrated, Wechsler Intelligence Scale for Children Fourth Edition Integrated (Wechsler *et al.*, 2004); WISC-IV, Wechsler Intelligence Scale for Children Fourth Edition (Wechsler, 2003).

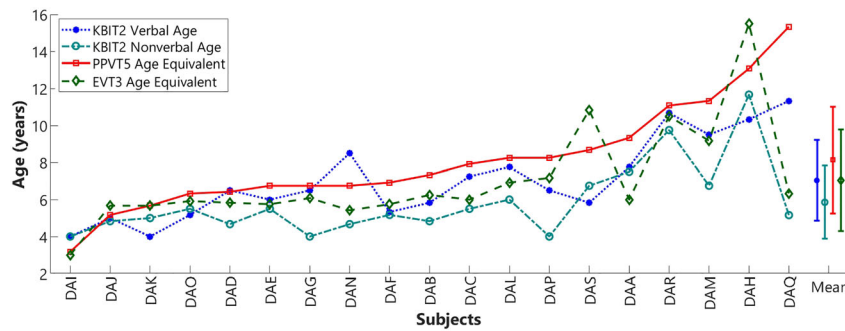


Figure 1. KBIT-2 verbal and non-verbal ages, as well as PPVT-5 and EVT-3 age equivalents in individuals with DS. Mean and standard deviation (SD, bars) for each age equivalent across participants are plotted at the far right. [Colour figure can be viewed at wileyonlinelibrary.com]

adjusted $P = .022$) and NEPSY-2 ($\rho = .737, P = .003$) but not with the EVT-3 ($\rho = .322, \text{adjusted } P = .210$). The Corsi Block Tapping Backward total score showed a significant positive correlation with the PPVT-5 ($\rho = .562, \text{adjusted } P = .027$) and non-significant correlations with the EVT-3 ($\rho = .388, \text{adjusted } P = .140$) and NEPSY-2 ($\rho = .421, \text{adjusted } P = .110$).

Discussion

Language is critical for adult quality of life as it shapes opportunities and experiences in employment, independent living and community engagement and social relationships. A substantial body of research has documented the language ability of children with DS (e.g. Abbeduto *et al.*, 2003; Lukowski *et al.*, 2019;

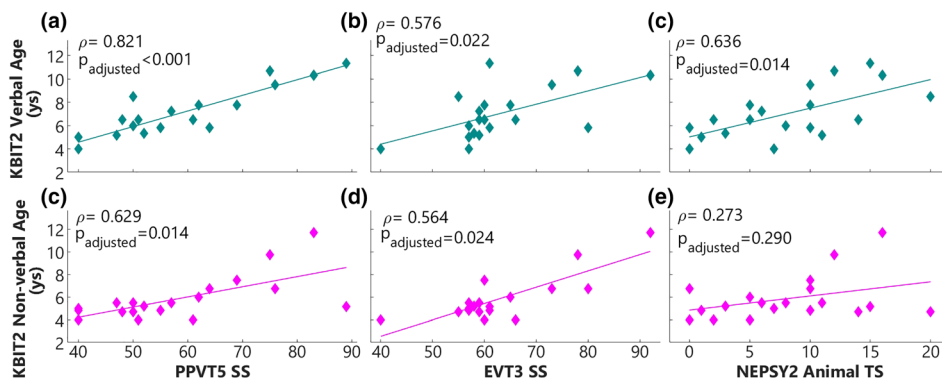


Figure 2. Spearman correlations between KBIT-2 verbal and non-verbal ages and language skills. The correlations between KBIT-2 verbal ages (green diamonds) and PPVT-5 and EVT-3 standard scores (SS) and the NEPSY-2 total scores (TS) in individuals with DS are shown in panels (a), (b) and (c), respectively. The correlations between KBIT-2 non-verbal ages (magenta diamonds) and the PPVT-5 and EVT-3 SS and the NEPSY-2 TS are shown in panels (d), (e) and (f), respectively. [Colour figure can be viewed at wileyonlinelibrary.com]

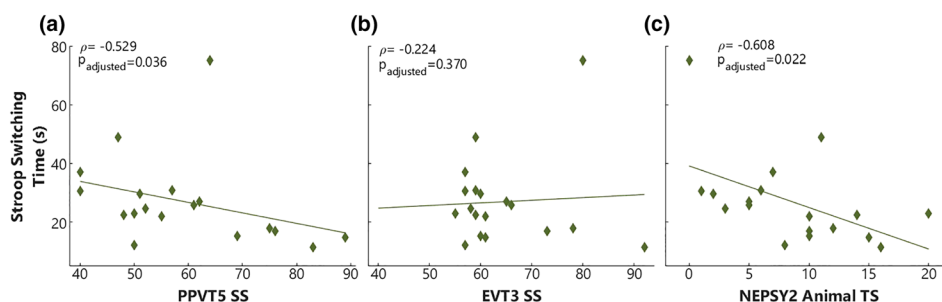


Figure 3. Spearman correlations between language skills and inhibition. The correlations between Cat and Dog Stroop switching time and the PPVT-5 standard scores (SS) and EVT-3 SS and the NEPSY-2 total scores (TS) are shown in panels (a), (b) and (c), respectively. [Colour figure can be viewed at wileyonlinelibrary.com]

Roberts *et al.*, 2007). However, much less is known about the language ability of young adults with DS. The current study provides valuable information about the language ability of young adults with DS and is also among the first to examine the connection between executive functioning and language in young adults with DS.

The first aim of the current study was to describe the language ability of young adults with DS on measures of receptive and expressive vocabulary and verbal fluency. On average, young adults with DS scored more than 2 SDs below the mean on measures of receptive and expressive vocabulary and demonstrated variability in ability to list animals during the verbal fluency task ($M = 8.61$, $SD = 5.53$.) In line with previous findings on children with DS (e.g. Abbeduto *et al.*, 2003; Martin *et al.*, 2009; Naess *et al.*, 2011), the

young adults with DS in the current study demonstrated larger delays in their expressive vocabulary (EVT-3) relative to their receptive vocabulary (PPVT-5). Thus, across childhood and into young adulthood, expressive language appears to be a relative weakness in DS. This finding builds on previous research describing persistent difficulties with expressive language for individuals with DS (e.g. Grieco *et al.*, 2015). Understanding language performance in young adults with DS is a critical first step to supporting future intervention studies. Information from the current study can be used to inform families and care teams about the range of baseline ability levels expected in young adults with DS using multi-word spoken phrases, advocate for individual-level assessment and supports and help gauge intervention-related improvements.

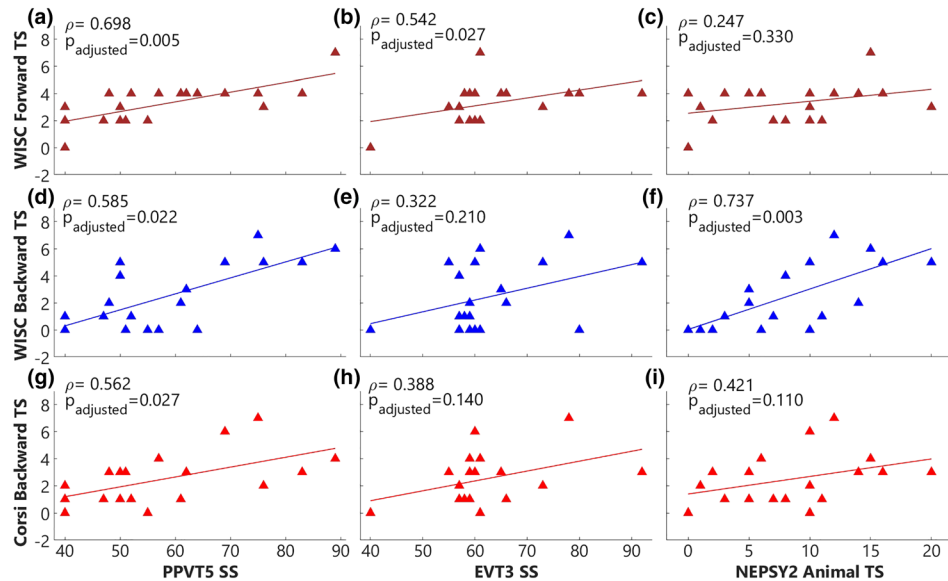


Figure 4. Spearman correlations between language skills and executive functioning. Panels (a), (d) and (g) show the correlations between PPVT-5 standard scores (SS) and total scores (TS) for WISC-IV Digit Span Forward (brown), WISC-IV Digit Span Backward (blue) and Corsi Block Tapping Task – Backward (red) in individuals with DS, respectively. The correlations between EVT-3 SS and the TS for the three tasks of executive functioning are shown in panels (b), (e) and (h). The correlations between NEPSY-2 TS and the total scores for the same three tasks are shown in panels (c), (f) and (i). [Colour figure can be viewed at wileyonlinelibrary.com]

The second aim of the current study was to evaluate the relationship between language measures and overall cognitive mental age. In contrast to our hypothesis, on average, the young adults with DS demonstrated receptive and expressive vocabulary skills that were higher than their overall non-verbal cognitive ability. This pattern is in contrast to findings for children with DS, which have often reported that expressive language is below that of overall cognitive ability (Abbeduto *et al.*, 2003; Martin *et al.*, 2009; Naess *et al.*, 2011). It is possible that our finding reflects that receptive and expressive vocabulary, the specific aspect of language captured in the PPVT-5 and EVT-3, is relatively well developed relative to overall cognitive ability for individuals with DS, whereas other aspects of expressive language (e.g. syntax) are not. Previous studies have indicated that syntax and utterance complexity are areas of particular weakness for individuals with DS across the lifespan (Martin *et al.*, 2009; Naess *et al.*, 2011). Interestingly, verbal fluency as assessed on the NEPSY-2 only had trend-level associations with overall cognitive ability (KBIT-2). This observation may mean that verbal fluency is not as strongly tied to overall cognitive ability as is vocabulary.

Future studies examining cognition in young adults with DS might consider that the KBIT-2 was somewhat limiting (i.e. one participant performed at floor level: <4 years, 0 months). The KBIT-2 may not be well suited to describe the cognitive abilities of young adults with DS with severe and profound ID.

The final aim of the current study was to determine the association between language and executive functioning skills in young adults with DS, and important connections between the two domains were identified. Specifically, there were strong positive associations between receptive vocabulary and inhibition, attention and verbal and non-verbal working memory. Young adults with DS who had larger receptive vocabularies demonstrated higher (vs. lower) inhibition, attention and verbal and non-verbal working memory. These findings are consistent with previous research relating receptive vocabulary with attention and inhibition for adults with DS (e.g. Faight & Connors, 2019; Laws & Gunn, 2004). Verbal fluency was associated with a smaller number of executive functioning skills, namely, inhibition and verbal working memory performance. Stavroussi *et al.* (2016) also highlighted a relationship between verbal working memory and verbal fluency skills for

adults with DS. The current study builds on previous research by showing that inhibition and verbal working memory in particular, rather than non-verbal working memory, may be critically related to verbal fluency for individuals with DS (e.g. Stavroussi *et al.*, 2016). Interestingly, only attention was associated with expressive vocabulary in the young adults with DS. If these associations are borne on future longitudinal and experimental studies, they could suggest that behavioural and medication efforts aimed at strengthened executive functioning in young adults with DS must be targeted to the specific domain of interest (e.g. targeting inhibition could feasibly be a way to improve receptive vocabulary and verbal fluency, but may not strengthen expressive vocabulary). Further research is needed to determine whether executive functioning skills offer benefits to other aspects of expressive language (e.g. grammatical markers) outside of expressive vocabulary, given that such associations have been reported elsewhere (e.g. Faught & Conners, 2019; Slevc, 2011).

The current study had both strengths and limitations. The study is among the few to describe language in young adults with DS and drew on standardised measures that are commonly used in other populations. Bonferroni correction was used to control for multiple comparisons. However, the study was limited by a relatively small sample size and did not include a control group (and instead drew on norms from the typically developing population). Of note, individuals with DS have wide variability in communication skills; given our inclusion criteria, the current study's findings may generalise most closely to young adults with DS who are using multi-word phrases. Measures of cognition and executive functioning also had limits. Floor effects and limited range were issues with the KBIT-2 and *WISC-IV* Digit Span Backward. In fact, the KBIT-2 age equivalent scale only goes down to 4 years, whereas the PPVT-5 and EVT-3 age equivalents go down to 2 years. Inclusion of a non-verbal measure of working memory supported ongoing efforts to better understand possible confounds involved when examining the relationship of verbal executive functioning tasks with language skills.

The study was also cross sectional, meaning that time-ordered interpretations about the connection between executive functioning and language in DS cannot be made. Although executive functioning is

thought to be a part of the foundation for language, there is also evidence that language fosters improvements in executive functioning. Thus, there is a need for longitudinal and experimental research to address the directionality of these connections.

In conclusion, the current study builds on the scant literature documenting language of young adults with DS and examining the potential role of executive functioning in shaping language ability. Our results suggest that young adults with DS exhibit receptive and expressive vocabularies generally commensurate with their overall non-verbal and verbal IQ. Inhibition, attention and verbal and non-verbal working memory had strong associations with receptive vocabulary, identifying important relationships for further investigation. Inhibition and verbal working memory were also associated with verbal fluency, whereas only attention was associated with expressive vocabulary.

These findings may suggest that interventions aimed at improving executive functioning, vocabulary and verbal fluency may be a meaningful pathway for enhancing daily living skills for young adults with DS. Indeed, there is evidence from populations without DS that medication and behavioural strategies (e.g. exercises aimed at training the ability to sustain attention and focus, inhibit impulses and engage in planful behaviour) are related to subsequent gains in language in children (e.g. Friedman & Sterling, 2019). Further research is needed to determine if similar approaches are useful in DS using both direct and caregiver-report assessment.

Acknowledgements

The authors thank the individuals with DS and parents who participated in this study, as well as GiGi's Playhouse Down Syndrome Achievement Center, Madison Area Down Syndrome Society and Waisman Center Clinical Translational Core team members who kindly aided in participant recruitment. The authors also thank members of the Binaural Hearing and Speech Lab for their help with data collection and coding.

Conflict of Interest

The authors have declared that no competing interests existed at the time of publication.

Source of Funding

This research was supported by NIH (Ro1 DCo03083 to Litovsky, U54 HD090256 to Chang, U19).

Data Availability Statement

The data that support the findings of this study are available from the corresponding author upon reasonable request.

References

- Abbeduto L., Murphy M. M., Cawthon S. W., Richmond E. K., Weissman M. D., Karadottir S. *et al.* (2003) Receptive language skills of adolescents and young adults with Down or fragile X syndrome. *American Journal on Mental Retardation* **108**, 149–60.
- Baddeley A. (2012) Working memory: Theories, models, and controversies. *Annual Review of Psychology* **63**, 1–29.
- Ball S. L., Holland A. J., Treppner P., Watson P. C. & Huppert F. A. (2008) Executive dysfunction and its association with personality and behaviour changes in the development of Alzheimer's disease in adults with Down syndrome and mild to moderate learning disabilities. *British Journal of Clinical Psychology* **47**, 1–29.
- Bishop D. V. M. & Norbury C. F. (2005) Executive functions in children with communication impairments in relation to autistic symptomology. I: Generativity. *Autism*, **9**, 7–27.
- Borella E., Carretti B. & Lanfranchi S. (2013) Inhibitory mechanisms in Down syndrome: Is there a specific or general deficit? *Research in Developmental Disabilities* **34**, 65–71.
- Carney D. P. J., Brown J. H. & Henry L. A. (2013) Executive function in Williams and Down syndromes. *Research in Developmental Disabilities* **34**, 46–55.
- Chapman R. S. & Hesketh L. J. (2000) Behavioral phenotype of individuals with Down syndrome. *Mental Retardation and Developmental Disabilities Research Reviews* **6**, 84–95.
- Costanzo F., Varuzza C., Menghini D., Addona F., Ganesini T. & Vicari S. (2013) Executive functions in intellectual disabilities: A comparison between Williams syndrome and Down syndrome. *Research in Developmental Disabilities* **34**, 1770–80.
- Dawson P. & Guare R. (2004) *Executive skills in children and adolescents: A practical guide to assessment and intervention*. Guilford Press, New York, New York.
- Dunn D. M. (2019) *Peabody Picture Vocabulary Test*. [Measurement Instrument], 5th edn. NCS Pearson, Bloomington, MN.
- Ellis Weismer S., Plante E., Jones M. & Tomblin J. B. (2005) A functional magnetic resonance imaging investigation of verbal working memory in adolescents with specific language impairment. *JSLHR* **48**, 405–25.
- Esbensen A. J., Hooper S. R., Fidler D., Hartley S. L., Edgin J., Liogier d'Ardhuy X. *et al.* (2017) Outcome measures for clinical trials in Down syndrome. *American Journal on Intellectual and Developmental Disabilities* **122**, 247–81.
- Faught G. G. & Conners F. A. (2019) Modeling the relations among sustained attention, short-term memory, and language in Down syndrome. *American Journal on Intellectual and Developmental Disabilities* **124**, 293–308.
- Friedman L. & Sterling A. (2019) A review of language, executive function, and intervention in autism spectrum disorder. *Seminars in Speech and Language* **40**, 291–304.
- Godfrey M. & Lee N. R. (2018) Memory profiles in Down syndrome across development: A review of memory abilities through the lifespan. *Journal of Neurodevelopmental Disorders* **10**, 1–31.
- Grieco J., Pulsifer M., Seligsohn K., Skotko B. & Schwartz A. (2015) Down syndrome: Cognitive and behavioral functioning across the lifespan. *American Journal of Medical Genetics Part C (Seminars in Medical Genetics)* **169C**, 135–45.
- Hamburg S., Rosch R., Startin C. M., Friston K. J. & Strydom A. (2019) Dynamic causal modeling of the relationship between cognition and theta-alpha oscillations in adults with Down syndrome. *Cerebral Cortex* **29**, 2279–90.
- Hartley S. L., Handen B. L., Devenny D. A., Mihaila I., Hardison R., Lao P. J. *et al.* (2017) Cognitive decline and brain amyloid- β accumulation across 3 years in adults with Down syndrome. *Neurobiology of Aging* **58**, 68–76.
- Hoyo L. D., Xicota L., Sanchez-Benavides G., Cuenca-Royo A., de Sola S., Langohr K. *et al.* (2015) Semantic verbal fluency pattern, dementia rating scores and adaptive behavior correlate with plasma Abeta42 concentrations in Down syndrome young adults. *Frontiers in Behavioral Neuroscience* **9**, 301.
- Kaufman A. S. & Kaufman N. L. (2004) *Kaufman brief intelligence test*. [Measurement Instrument]. 2nd edn. AGS Publishing, Circle Pines, MN.
- Kaushanskaya M., Park J., Gangopadhyay I., Davidson M. & Ellis Weismer S. (2017) The relationship between executive functions and language abilities in children: A latent variables approach. *JSLHR* **60**, 912–23.
- Khanna M. M. & Boland J. E. (2010) Children's use of language context in lexical ambiguity resolution. *The Quarterly Journal of Experimental Psychology* **63**, 160–93.
- Korkman M., Kirk U. & Kemp S. (2007) NEPSY—Second Edition [Measurement Instrument]. San Antonio, TX: Harcourt Assessment.
- 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.

- Laws G. & Gunn D. (2004) Phonological memory as a predictor of language comprehension in Down syndrome: a five-year follow-up study. *Journal of Child Psychology and Psychiatry* **45**, 326–37.
- Lukowski A. F., Milojevich H. M. & Eales L. (2019) Cognitive functioning in children with Down syndrome: Current knowledge and future directions. *Advances in Child Development and Behavior* **56**, 257–89.
- Lynch K. M., Shi Y. G., Toga A. W., Clark K. A. & Neurocognition P. I. (2019) Hippocampal shape maturation in childhood and adolescence. *Cerebral Cortex* **29**, 3651–65.
- Martin G. E., Klusek J., Estigarribia B. & Roberts J. E. (2009) Language characteristics of individuals with Down syndrome. *Topics in Language Disorders* **29**, 112–32.
- Marton K. (2008) Visuo-spatial processing and executive functions in children with specific language impairment. *International Journal of Language and Communication Disorders* **43**, 181–200.
- Miyake A., Friedman N. P., Emerson M. J., Witzki A. H., Howerter A. & Wager T. D. (2000) The unity and diversity of executive functions and their contributions to complex "frontal lobe" tasks: A latent variable analysis. *Cognitive Psychology* **41**, 49–100.
- Naess K. A. B., Lyster S. A. H., Hulme C. & Melby-Lervag M. (2011) Language and verbal short-term memory skills in children with Down syndrome: A meta-analytic review. *Research in Developmental Disabilities* **32**, 2225–34.
- O'Hare E. D. & Sowell E. R. (2008) Imaging developmental changes in gray and white matter in the human brain. In: *Handbook of developmental cognitive neuroscience* (eds A. N. Charles & M. Luciana), 2nd edn, pp. 23–38. Cambridge, MA: MIT Press.
- Pennington B. F., Moon J., Edgin J., Stedron J. & Nadel L. (2003) The neuropsychology of Down syndrome: Evidence for hippocampal dysfunction. *Child Development* **74**, 75–93.
- Roberts J. E., Price J. & Malkin C. (2007) Language and communication development in Down syndrome. *Mental Retardation and Developmental Disabilities Research Reviews* **13**, 26–35.
- Rowe J., Lavender A. & Turk V. (2006) Cognitive executive function in Down's syndrome. *The British Psychological Society* **45**, 5–17.
- Seung H. K. & Chapman R. (2000) Digit span in individuals with Down syndrome and in typically developing children: Temporal aspects. *Journal of Speech, Language, and Hearing Research* **43**, 609–20.
- Slevc L. R. (2011) Saying what's on your mind: Working memory effects on sentence production. *Journal of Experimental Psychology: Learning, Memory, and Cognition* **37**, 1503–14.
- Stavroussi P., Andreou G. & Karagiannopoulou D. (2016) Verbal fluency and verbal short-term memory in adults with Down syndrome and unspecified intellectual disability. *International Journal of Disability, Development and Education* **63**, 122–39.
- Traverso L., Fontana M., Usai M. C. & Passolunghi M. C. (2018) Response inhibition and interference suppression in individuals with Down syndrome compared to typically developing children. *Frontiers in Psychology*, 1–14.
- Vugs B., Hendriks M., Cuperus J. & Verhoeven L. (2014) Working memory performance and executive function behaviors in young children with SLI. *Research in Developmental Disabilities* **35**, 62–74.
- Wechsler D. (2003) *Wechsler intelligence scale for children*. 4th edn, PsychCorp, San Antonio, TX.
- Wechsler D., Kaplan E., Fein D., Kramer J., Morris R., Delis D., & Maerlender, A. (2004) *Wechsler intelligence scale for children integrated [Measurement Instrument]*, 4th edn. Harcourt, San Antonio, TX.
- Williams K. T. (2019) *Expressive vocabulary test [Measurement Instrument]*, 3rd edn. NCS Pearson, Bloomington, MN.
- Witecy B. & Penke M. (2017) Language comprehension in children, adolescents, and adults with Down syndrome. *Research in Developmental Disabilities* **62**, 184–96.
- Woodard K., Pozzan L. & Trueswell J. C. (2016) Taking your own path: Individual differences in executive function and language processing skills in child learners. *Journal of Experimental Child Psychology* **141**, 187–209.
- Yang Y., Conners F. A. & Merrill E. C. (2014) Visuo-spatial ability in individuals with Down syndrome: Is it really a strength? *Research in Developmental Disabilities* **35**, 1473–500.

Accepted 14 June 2021