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Evaluation of executive functioning in people with intellectual disabilities

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Abstract

Background Executive functioning (EF) is an important concept in cognitive psychology that has rarely been studied in people with intellectual disabilities (IDs). The aim of this study was to examine the validity of two test batteries and the structure of EF in this client group.
Methods We administered the children's version of the Behavioural Assessment of the Dysexecutive Syndrome (BADS-C) and the Cambridge Executive Functioning Assessment (CEFA) for people with ID, to 40 participants who attended day centres for people with mild to moderate learning disabilities [mean full-scale intelligence quotient (IQ) = 59]. The BADS-C consists of six EF subtests while the CEFA contains eight EF (including two executive memory) subtests and four memory subtests. IQ and receptive language ability were also assessed. The results were subjected to principal components analysis, and regression analysis was used to examine the relationship of the ensuing factors to other cognitive variables.
Results Scores on both sets of EF tests were only weakly related to receptive language ability, and even more weakly related to IQ. Scores on the BADS-C were substantially lower than predicted from the published norms for people in higher IQ ranges, and many participants scored zero on three of the six subtests. This potential floor effect was less evident with scores on the CEFA. Principal components analyses produced one usable factor for the BADS-C, and two factors for the CEFA that differed in both the extent of involvement of working memory and the predominant sensory modality. A combined analysis of the subtests retained from both analyses produced three factors that related uniquely to aspects of IQ and memory.
Conclusions The CEFA is suitable for use with people with mild to moderate learning disabilities, whereas the BADS-C is at the lower limit of usability with this client group. The lower-than-expected scores observed on the BADS-C may indicate that people known to learning disability services may be more impaired than people of comparable IQ not known to services. The structure of EF seen in people with IDs closely resembles a model of EF in the general population that has received a broad level of support.

Keywords Behavioural Assessment of the Dysexecutive Syndrome, Cambridge Executive Functioning Assessment, executive functioning, intellectual disability, IQ, receptive language ability

Introduction

Executive functioning (EF) refers to the set of abilities involved in planning, self monitoring and purposive action, which, it has been argued, are 'at the heart of all socially useful, personally enhancing,

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constructive and creative activities' (Lezak 1982). Many cognitive processes underpin EF, including response initiation, response inhibition, attention, working memory, set shifting and fluency (see e.g. Pennington & Ozonoff 1996; Manly & Robertson 1997; Busch *et al*. 2005a; Rowe *et al.* 2006). Numerous neuropsychological tests purport to assess aspects of EF, and a number of batteries have been developed that combine tests in order to sample several of these putative underlying processes: examples include the Behavioural Assessment of the Dysexecutive Syndrome (BADS: Wilson *et al.* 1996), the Delis–Kaplan Executive Function System (Delis *et al.* 2004) and the Complex Task Assessment System (Wolf *et al.* 2008). The diversity of the concepts discussed in relation to EF has stimulated debate over whether EF can be considered as unitary (e.g. Duncan *et al.* 2000), or multidimensional (e.g. Andrez & van der Linden 2002), or something in between, involving a set of distinct processes with some over-arching coordination (e.g. Robbins *et al.* 1998; Miyake *et al.* 2000).

There has been little investigation of EF in people with intellectual disabilities (IDs), and nothing is known about the extent to which EF is a unitary or a multidimensional construct in this context. There is a small literature in some specific developmental disorders, such as Down syndrome (e.g. Das *et al.* 1995; Pennington *et al.* 2003; Rowe *et al.* 2006; Ball *et al.* 2008), Prader–Willi syndrome (e.g. Gross-Tsur *et al.* 2001; Walley & Donaldson 2005; Jauregi *et al.* 2007; Woodcock *et al.* 2009) and fragile X syndrome (e.g. Wilding *et al.* 2002; Cornish *et al.* 2009; Woodcock *et al.* 2009). With the notable exception of some of the studies in Down syndrome, these studies have typically used a narrow range of tests, targeted, often incidentally, at specific aspects of EF, rather than a comprehensive test battery. When broader test batteries have been used (e.g. Pennington *et al.* 2003; Walley & Donaldson 2005; Rowe *et al.* 2006; Ball *et al.* 2008), these are never standardised across studies or directly comparable to the test batteries used with the general population.

There are two approaches to test development for people with IDs: to adapt tests developed and validated for more able people, sometimes using children's versions of adult tests, or to develop new tests that are specifically designed to be more accessible. In this study we have examined two test batteries that exemplify each of these approaches. The children's version of the BADS (BADS-C: Emslie *et al.* 2003) is a simplified version of the adult BADS, validated on children from age 8 and IQ range 70–90. The BADS-C is used clinically in some learning disability services, but we are unaware of any literature on the subject. The CEFA (Ball *et al.* 2008) is a battery of EF and memory tests that was developed for use in the detection of early dementia in people with learning disabilities and Down syndrome (Ball *et al.* (2008).

In the present study we administered both the BADS-C and the CEFA to a population of people with mild learning disabilities of mixed aetiology. Our aims were to:

1 Evaluate the utility of these test batteries for people with learning disabilities;

2 Establish the extent to which each of the batteries measures a unitary construct of EF; and

3 Investigate whether the BADS-C and the CEFA measure the same underlying constructs.

Method

Participants

The participants were 40 people [52.5% male, with a mean [standard deviation (SD)] age of 40.1 (10.8) years: range, 20–63], who attended day centres for people with mild to moderate learning disabilities. All participants provided informed consent and the study was approved by the Local National Health Service Research Ethics Committee. Participants' disabilities were heterogeneous: no attempt was made to record aetiology, which in many cases would be unknown.

Cognitive ability

Participants were assessed for intellectual ability using the Wechsler Abbreviated Scale of Intelligence (WASI), and for receptive language ability using the British Picture Vocabulary Scale (2nd edition) (BPVS). The WASI Verbal IQ (VIQ) and Performance IQ (PIQ) scores are equivalent to the Verbal Comprehension Index and the Perceptual Organization Index on the Wechsler Adult Intelligence Scale (WAIS), and are considered to provide 'purer' measures of intellectual ability than the WAIS VIQ and PIQ scores, which also include elements of EF. Participants' mean (SD) scores were: Full-Scale IQ, 59.1 (5.1), range 50–70; BPVS raw score, 80.4 (27.3): age equivalent of mean raw score, 7:10.

Executive functioning

The BADS-C (Emslie *et al.* 2003) consists of six tests of EF. The CEFA (Ball *et al.* 2008) includes six EF tests (CEFA-EF) and six memory tests (CEFA-M), two of which (Object Memory and Prospective Memory) are considered to represent 'executive memory'. Brief descriptions of each task are shown in Table 1. Full details may be found in Emslie *et al.* (2003) and Ball *et al.* (2008).

Procedure

After first obtaining informed consent, which was witnessed by a member of the day-centre staff, tests were conducted over five sessions of approximately 1 h each. The WASI and BPVS were administered in session 1, the BADS-C in session 4 and the CEFA in session 5. In sessions 2 and 3 other tests were administered that will be reported elsewhere. After each session, service users were asked to rate their experience of the session on a 3-point scale ('I didn't like it'; 'I liked it a bit'; 'I liked it a lot'). Of the 120 sessions, 112 were rated as 'liked a lot', 8 as 'liked a bit' and none as 'not liked'.

The procedure for the BADS-C was as described in the user guide (Emslie *et al.* 2003) with some minor modifications. In addition to the written versions, the rules for the Playing Cards and Six-Part Tests were also provided in a visual format. Also, in the Six-Part Test, participants were asked to make their responses verbally rather than in writing. These modifications were made in order to increase accessibility and decrease dependence on literacy. As the BADS-C has not been standardised on either adults or people with IQ < 70, scores on each subtest were converted to standardised scores using data for the oldest age group (15:00–15:11) and the lowest IQ range (70–90) for whom standardised scores are available (Emslie *et al.* 2003).

As described by Ball *et al.* (2008), the CEFA included administration of the Cambridge Cognitive Examination (CAMCOG: Roth *et al.* 1988) and the Severe Impairment Battery (SIB: Saxton *et al.* 1993), from which relevant memory items were extracted to find Immediate Memory, Delayed Recognition and Delayed Recall Composite Scores (see Ball *et al.* 2008 for details). In the present study, only those items from the CAMCOG and SIB scored by Ball *et al.* (2008) were presented, so that the entire CEFA could be administered in less than 1 h. Also, as the participants had by this time met the experimenter on several occasions, the item 'Recall of the experimenter's name' was replaced by the item 'Recall of a name – John Brown'. Otherwise, the CEFA was administered and scored as described by Ball *et al.* (2008).

Statistical analysis

Spearman rank-order correlations were used to examine relationships between individual EF or memory subtests and IQ or BPVS scores, because for several subtests, a high proportion of the participants had minimum or maximum scores. Because of the number of correlations computed, the probability level for significance was set at $P < 0.01$. *t*-tests were used to compare the present data with the published data of Ball *et al.* (2008); non-parametric tests could not be used for this purpose because the raw data were not available.

Performance on EF tests was analysed by principal components analysis, with varimax rotation. The data were also explored with the other orthogonal (equamax, quartimax) and oblique (oblimin, promax) rotations available in SPSS-16: all analyses produced identical factor structures with all rotations. Because there were insufficient participants to enter all of the experimental tasks into a single analysis, separate analyses were conducted on subsets of tasks, maintaining a minimum ratio of ≥5 cases per item (Tabachnik & Fidell 2006).

Stepwise multiple regression was used to examine the relationships between the factors identified and other cognitive variables, specifically: receptive language ability (BPVS); VIQ and PIQ; and, from the CEFA, two tests of immediate memory (Memory for Sentences and Immediate Memory) and two tests of delayed memory (Delayed Recall and Delayed Recognition). All variables were entered in a single block.

Table 1 Summary description of BADS-C and CEFA subtests*

Behavioural assessment of the dysexecutive syndrome – children's version (BADS-C)	
Playing cards	A series of playing cards are shown. After responding 'Yes to red; No to black' the rule changes to 'Say Yes to same as previous; No to different'. The second phase of the test is scored. The rule is visible throughout. The task requires the participant to remember the response to the previous card and to inhibit use of the rule learned in phase 1.
Water test	The task is to remove a cork from a tube, using the equipment provided to introduce water into the tube to float the cork, by making a series of steps in the correct order. If the participant fails to make the next step prompts are provided and points are lost. Points are also lost for perseverative responding.
Key search	Participant are shown a white square that represents a field where they have 'lost their keys'. The task is to draw a line to show how they would search the field to be sure of finding the keys. Points are gained for demonstrating a strategy that would cover the ground efficiently.
Zoo map 1	The participant is shown a map of a zoo and asked to trace out a continuous route that would be taken in order to visit various animals and places, with a restriction on retracing some of the paths. The list of attractions to visit is visible throughout. Points are gained for visits made in the correct order, and lost for breaking rules.
Zoo map 2	The test is repeated but this time the order of visits is specified, so there is no planning requirement.
Six-part test	Over 5 min, three tasks, each in two halves, must be attempted, with a restriction that the two halves of each task cannot be attempted sequentially. Points are gained for following the sequencing rule and allocating time to each of the tasks.
Cambridge executive functioning assessment: executive functioning tests (CEFA-EF)	
Cats and dogs	Participants are asked to name pictures of a 'cat' and a 'dog'. They are then asked to say 'cat' when shown a picture of a dog, and vice versa. Points are gained for correct performance in the second phase of the test. *(Involves response inhibition and working memory for rule maintenance.)*
Spatial reversal	A coin is hidden under one of two boxes. After the participant has learned to choose the correct box, the location is reversed. The reversal phase is scored. In order to score points the participant must detect the changed contingencies and respond accordingly.
Weigl sorting	Participants first sort coloured shapes into piles 'that go together'. They are then asked to 'make the piles differently'. The shift phase is scored. Prompts are given, and points lost, if the participant is unable to find an alternative sort. *(Involves extra-dimensional set shifting.)*
Tower of London	The apparatus consists of three coloured rings on three pegs. The task is to move the rings one at a time to configure them to a pattern demonstrated by the tester. A series of problems is presented graded in difficulty. The task requires a sequence of steps to be planned in advance of the first move. *(Involves planning & working memory.)*
Scrambled boxes	A coin is hidden under each of three or six visually distinct boxes. Points are gained for locating the coins. After each coin is found all of the boxes are moved to different positions. *(Involves working memory and response inhibition.)*
Verbal fluency	Points are scored for naming as many animals as possible within 1 min. *(Involves initiation, set shifting, working memory, and efficient organisation of retrieval and recall.)*
Cambridge executive functioning assessment: memory tests (CEFA-M)	
Object memory	One of a set of objects, that increases up to a maximum of six, is covered by a box. Points are scored for naming the hidden object. *(Involves cued retrieval and working memory.)*
Prospective memory	About 20 min before the end of the session the tester hides a set of keys. Points are scored for reminding the tester of this, and where they are, when the session ends.
Memory for sentences	The task is to repeat sentences that increase in length.
Immediate memory	This test includes a variety of recall and recognition items taken from the CAMCOG and the SIB.
Delayed recognition	Delayed recognition of two SIB objects and six CAMCOG pictures.
Delayed recall	Delayed recall of pictures and various verbal items, taken from the CAMCOG.

* For some of the subtests, the underlying processes (as proposed by Ball *et al.* 2008) are shown in italics; this information is referred to in the Discussion.
CAMCOG, Cambridge Cognitive Assessment; SIB, Severe Impairment Battery.

Results

Descriptive statistics

Table 2 shows descriptive statistics for the 18 individual BADS-C and CEFA subtests. A high proportion of participants (87.5%) scored 0 on the BADS Zoo Map 2 Test and relatively high proportions scored 0 on two other BADS subtests, Playing Cards (37.5%) and Zoo Map 1 (30%), as well as a single CEFA subtest, Spatial Reversal (42.5%). Conversely, a relatively high proportion of participants had maximum scores on the CEFA-EF subtests Weigl Sorting (32.5%) and, to a lesser extent, Cats and Dogs (25%). Similarly, in the CEFA-Memory battery, maximum scores were obtained by a high proportion of participants on Delayed Recognition (70%), and relatively high proportions of participants on Prospective Memory (32.5%) and Immediate Memory (45%).

Table 2 also shows the correlations of each subtest with BPVS and WASI scores. Significant correlations with BPVS scores were found for only two BADS subtests (Water Test and Zoo Map 1) and, more weakly, two CEFA-EF subtests [Tower of London and Verbal Fluency]. BPVS scores were more strongly related to performance on the CEFA-M tests, with significant correlations on five of the six subtests. Correlations with WASI scores were even weaker, with only one EF and three memory tests showing significant relationships. Correlations were also calculated for a composite memory score (the total of the two immediate and

Table 2 Descriptive statistics for the BADS-C and CEFA subtests

Test	Maximum score	Range of scores	No. at minimum (%)	No. at maximum (%)	Mean (SD)	Median	Correlation with BPVS	Correlation with WASI
Behavioural assessment of the dysexecutive syndrome – children's version (BADS-C)								
Playing cards	13	0–6	15 (37.5)	0 (0.0)	1.6 (1.5)	2	0.25	0.05
Water test	14	0–14	1 (2.5)	5 (12.5)	7.2 (3.3)	7	0.52**	0.26
Key search	16	0–6	3 (7.5)	0 (0.0)	4.6 (1.6)	4.5	0.06	0.11
Zoo map 1	13	0–8	12 (30.0)	0 (0.0)	3.0 (2.6)	3	0.48**	0.21
Zoo map 2	10	0–10	35 (87.5)	1 (2.5)	0.4 (1.6)	0	0.07	0.09
Six part test	13	2–10	0 (0.0)	0 (0.0)	3.5 (1.6)	3	–0.02	–0.03
Cambridge executive functioning assessment: executive functioning tests (CEFA-EF)								
Cats and dogs	16	0–16	2 (5.0)	10 (25.0)	10.6 (5.3)	12	0.22	0.10
Spatial reversal	7	0–6	17 (42.5)	0 (0.0)	2.6 (2.6)	2	0.23	0.14
Weigl sorting	5	0–5	3 (7.5)	13 (32.5)	2.8 (1.8)	2	0.27	0.47**
Tower of London	12	1–11	0 (0.0)	0 (0.0)	6.8 (2.6)	7	0.42*	0.13
Scrambled boxes	11	0–11	1 (2.5)	2 (5.0)	5.8 (2.3)	5.5	0.34	0.04
Verbal fluency	5	0–5	1 (2.5)	1 (2.5)	2.8 (1.1)	3	0.42*	0.36
Cambridge executive functioning assessment: memory tests (CEFA-M)								
Object memory	10	2–10	0 (0.0)	1 (2.5)	6.3 (2.0)	6	0.39*	–0.22
Prospective memory	4	0–4	1 (2.5)	13 (32.5)	2.7 (1.2)	3	0.51**	0.41*
Memory for sentences	49	0–48	2 (5.0)	0 (0.0)	34.5 (13.6)	39	0.73**	0.50**
Immediate memory	13	7–13	0 (0.0)	18 (45.0)	12.0 (1.5)	12	0.64**	0.29
Delayed recognition	8	3–8	0 (0.0)	28 (70.0)	7.4 (1.3)	8	0.61**	0.40*
Delayed recall	14	2–12	0 (0.0)	0 (0.0)	6.9 (2.4)	7	0.35	0.02

* $P < 0.01$, ** $P < 0.001$.

The first six columns show descriptive statistics for each of the subtests in the BADS-C and CEFA: the maximum score obtainable for each test and the range of scores obtained, the numbers of participants obtaining minimum and maximum scores, and the mean (standard deviation) and median scores. The final two columns show the relationships (Spearman correlation coefficients) between each variable and measures of receptive language ability (BPVS) and IQ (WASI).

BPVS, British Picture Vocabulary Scale; WASI, Wechsler Abbreviated Scale of Intelligence.

two delayed memory tests). The pattern of significant and non-significant results for the other eight subtests was identical to that for the BPVS (not shown); and there was a strong correlation between the BPVS and composite memory scores ($r = 0.68$, $P < 0.001$).

Comparisons with normative data

Children's version of the Behavioural Assessment of the Dysexecutive Syndrome

The BADS-C manual (Emslie *et al.* 2003) provides tables for three IQ ranges, 70–90, 90–110 and 110–130. Inspection of the tables shows that the difference in EF scores between the three IQ cohorts is relatively small. The table for IQ range 70–90 was used to look up the standardised score equivalents, for each subtest, of the raw scores corresponding to the 50th percentile of the population for each of the three IQ ranges. This showed that the overall improvement over the three IQ ranges is around 3–4 points on the BADS-C per 20 IQ points. On this basis, the present cohort, with IQs in the 50–70 range, would be predicted to have a standardised score of around 56, towards the bottom of the 'average' range relative to the IQ 70–90 cohort. In fact, the mean score of the present cohort on the BADS-C was 20 (Fig. 1), reflecting a severe impairment. This score is far below the bottom of the published scale (a score of 39), and well down within the lowest 0.1% of the normative sample of people with IQs in the 70–90 range.

Three potential explanations of this discrepancy were explored.

1 Scores on the BADS-C were referred to the table of norms for 8-year-olds (approximating to the mean age-equivalent of the present cohort on the BPVS). This has the effect of raising the mean score of the present cohort, but only to the middle of the 'borderline' range, equivalent to the 5th percentile.

2 The possibility of a general fall in cognitive abilities between the ages of 15 (the published norms) and 40 (the mean age of the present sample) was evaluated using the WASI manual (which covers age ranges between 6 and 89 years), by calculating the IQ scores that would be obtained if a 15-year-old produced the raw scores that generate a Full-Scale IQ score of 70 in a 40-year-old. A FSIQ score of 70 results from a *T*-score of 31 on each of the four WASI subtests. The raw scores producing this *T*-score were read off the score table for age 35–45, and the *T*-scores corresponding to these raw scores were read off the score table for age 15:8–15:11. These *T*-scores were then used to calculate IQ scores. This showed that, for IQs around 70, verbal ability increases by a few points between age 15 and age 40, while non-verbal ability decreases by a few points, with a net 1-point decrease in FSIQ score from 70 to 69.

3 The insert in Fig. 1 shows the breakdown of BADS-C scores across the IQ range 50–70. This demonstrates that there is no steep drop-off in BADS-C performance below an IQ of 70. The trend line identifies a 9-point fall in BADS-C score

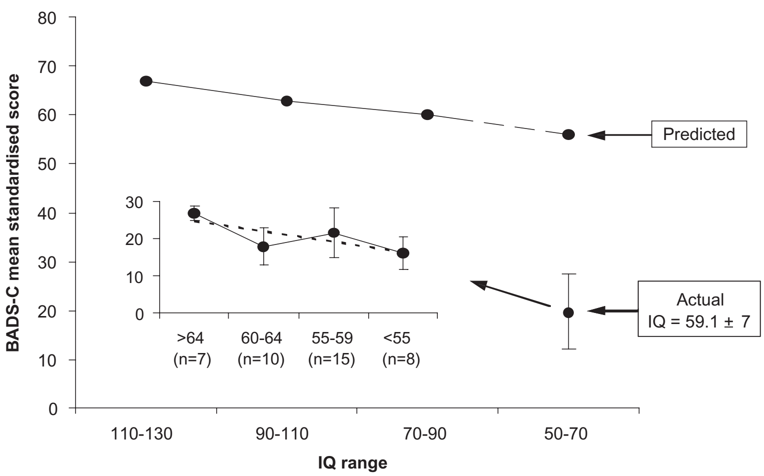

Figure 1 The main part of the figure shows the predicted children's version of the Behavioural Assessment of the Dysexecutive Syndrome (BADS-C) score for people in the IQ range 50–70, based on the normative scores for higher IQ ranges, and the actual scores of the present cohort (mean ± standard deviation). The inset shows the actual scores (mean ± standard deviation) broken down into 5-point IQ ranges, with the trend line.

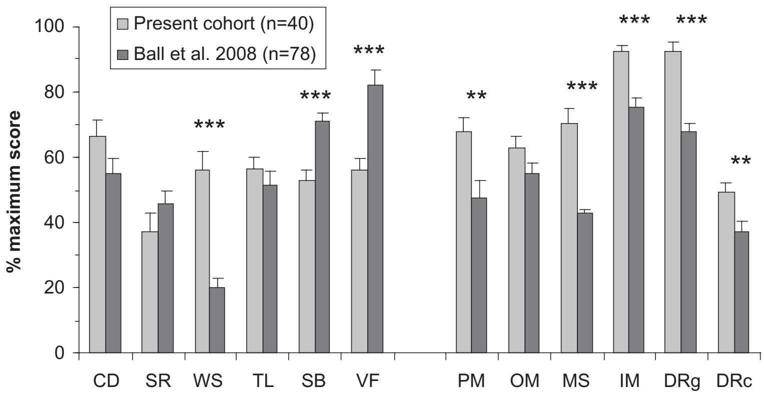

Figure 2 Scores on the 12 Cambridge executive functioning assessment for people with intellectual disability subtests (executive functioning tests to the left and memory tests to the right), comparing the present data with those of Ball *et al.* (2008). Score are expressed as the % of the maximum score on each test (mean + standard error). $^{**}P < 0.01$, $^{***}P < 0.001$. See text for identification of the tests.

across the 20-point IQ range, only a little greater (relative to the 35-point difference between actual and predicted scores) than the 3–4 BADS-C/20 IQ point fall at the higher IQ ranges.

Cambridge Executive Functioning Assessment for people with intellectual disability

Figure 2 compares the scores of the present cohort with those reported by Ball *et al.* (2008) for their cohort of people with Down syndrome (but without signs of dementia). The present cohort were significantly more able than the Down syndrome cohort, who had a mean (SD) BPVS score of 55.0 (27.3), compared with the mean for the present sample of 80.4 (27.3) ($t = 4.79$, $P < 0.001$). A comparison of the present data with Table 3 of Ball *et al.* (2008) shows that the present sample also had higher scores on all of the CEFA memory subtests, and on all but one subtest (Object Memory) these differences were significant.

In contrast to the superior performance of the present sample on memory tests, the two samples were almost identical in their performance on the CEFA executive functioning battery (mean total scores: 31.4 vs. 31.1). However, there were some differences in the profile of strengths and weaknesses, with the present cohort scoring significantly higher on one subtest, Weigl Sorting, and the Down Syndrome cohort scoring significantly higher on two subtests, Scrambled Boxes and Verbal Fluency.

Factor structure

Principal components analysis of the BADS-C produced two factors, accounting for 58% of the variance (Table 3). Two subtests – Zoo Map 2 and the Six-Part Test – loaded highly on Factor 1; three subtests – Playing Cards, Water Test and Zoo Map 1 – loaded highly on Factor 2; one subtest, Key Search did not contribute strongly to either factor. Factor 2 was normally distributed (low skew and kurtosis and a non-significant Shapiro–Wilks test: Table 3), but Factor 1 was not. This, combined with the high proportion of participants scoring at ceiling on one of the two contributing subtests (Zoo Map 2), suggests that Factor 1 cannot be considered valid. It will not be considered further.

A second principal components analysis was conducted on eight CEFA subtests: the six subtests in the EF battery and the two Executive Memory Tests (Table 3). This analysis also produced two factors, accounting for 55% of the variance. Three subtests loaded highly on Factor 1: Tower of London, Scrambled Boxes and Object Memory. Four subtests loaded highly on Factor 2: Cats and Dogs, Weigl Sorting Verbal Fluency and Prospective Memory. The eighth CEFA subtest, Spatial Reversal, was more strongly associated with Factor 1, but with a relatively low loading. Both factors were normally distributed.

Scores on BADS-C Factor 2 did not correlate significantly with either Factor 1 ($r = 0.24$) or Factor 2 ($r = 0.30$) of the CEFA. In order to explore these relationships further (while maintaining a ratio >5 of cases to items), two further principal components analyses were run in which the subtests with high loadings on BADS-C Factor 2 were entered alongside the subtests with high loadings on CEFA Factor 1 or Factor 2 (results not

Table 3 Principal components analysis of the BADS-C and CEFA

A: BADS-C	Factors F1	Factors F2	B: CEFA	Factors F1	Factors F2
Eigenvalue	2.13	1.35	Eigenvalue	2.24	1.09
% Variance	35.53	22.45	% Variance	37.36	18.16
Cumulative %	35.53	57.98	Cumulative %	37.36	55.15
Item loadings			Item loadings		
Playing cards	0.33	**0.61**	Cats and dogs	0.42	**0.62**
Water test	–0.43	**0.76**	Spatial reversal	0.46	–0.05
Key search	0.33	0.26	Weigl sorting	–0.19	**0.64**
Zoo map 1	0.23	**0.64**	Tower of London	**0.66**	0.29
Zoo map 2	**0.87**	0.13	Scrambled boxes	**0.75**	0.23
Six-part test	**0.90**	0.06	Verbal fluency	0.14	**0.77**
			Object memory	**0.78**	0.07
			Prospective memory	0.30	**0.69**
Normality tests					
Skewness	3.38	0.45		–0.46	–0.14
Kurtosis	14.97	–0.55		–0.82	–0.75
Shapiro–Wilks	0.664	0.959		0.974	0.966
	$P < 0.001$	$P = 0.16$		$P = 0.46$	$P = 0.27$

* Items loading at >0.5 are highlighted in bold.
BADS-C, behavioural assessment of the dysexecutive syndrome – children's version; CEFA, Cambridge executive functioning assessment.

shown). In each of these analyses, the CEFA subtests loaded onto a single factor and the BADS-C Water Test and Playing Cards tests loaded onto a second factor (designated Combined Factor 3, to avoid confusion with CEFA Factor 2). However, the BADS-C Zoo Map 1 subtest loaded (at 0.63) alongside the CEFA Factor 1 subtests rather than the other BADS-C subtests.

Regression analyses were used to assist in the interpretation of the factor scores. These analyses (Table 4) showed that the different factors had unique relationships with other cognitive variables.

1 Both CEFA Factor 1 and Combined Factor 1 (which included the same subtests as CEFA Factor 1 plus the BADS Zoo Map test) were associated with Delayed Recognition;

2 The CEFA Factor 2 was associated with Immediate Memory and VIQ; and

3 Both BADS Factor 2 and Combined Factor 3 (which comprised two of the three BADS Factor 2 subtests) were associated with receptive language ability (BPVS).

Discussion

Relationship of Executive functioning to IQ

Significant relationships between IQ and EF have been reported in intellectually more able populations, but the correlations are low (around 0.3), accounting for less than 10% of the variance (Ardila *et al.* 2000; Duff *et al.* 2005). Some components of IQ relate more closely to EF than do others (Duncan *et al.* 1995), and vice versa (Friedman *et al.* 2006). Previous work on EF in people with IDs includes studies that report similar IQ but different EF (Rowe *et al.* 2006), similar EF but different IQ (Maehler & Schuchardt 2009), and no intra-group correlation between these two variables (Jauregi *et al.* 2007).

Ball *et al.* (2008) reported that on most CEFA subtests, performance of people with Down syndrome was significantly related to receptive language ability (BPVS scores) but this finding was only partly supported by the present data in a heterogeneous sample. Strong correlations were found with CEFA-M tests, but only one CEFA-EF

Table 4 Results of regression analyses relating executive functioning factor scores with cognitive variables

Factor	CEFA F1	CEFA F2	BADS F2	Combined F1	Combined F3
Subtests	Tower of London Scrambled boxes Object memory	Cats and dogs Verbal fluency Prospective memory Weigl sorting	Playing cards Water test Zoo map 1	Cefa Factor 1 zoo map 1	Playing cards Water test
F, r^2	5.82*, 0.11	13.37***, 0.39	22.4***, 0.35	11.17***, 0.21	10.11***, 0.19
British picture vocabulary scale			$t = 4.73$***	$t = 3.29$***	$t = 3.18$***
Verbal intelligence quotient		$t = 3.18$***			
Immediate memory		$t = 2.72$**			
Delayed recognition	$t = 2.42$*				

* $P < 0.05$, ** $P < 0.01$, *** $P < 0.005$.
The first three columns show the significant relationships resulting from regressions of the variables in the first column (as well as performance IQ, Memory for Sentences and Delayed Recall) onto each of the factors identified in Table 3. The final two columns show the same analysis in relation to two factors emerging from a combined analysis of the CEFA Factor 1 and BADS-C Factor 2, in which the BADS-C subtests split between the two factors (see text). The factors are designated 'Combined Factor 1' and 'Combined Factor 3' to avoid confusion with CEFA Factor 2.
BADS-C, Behavioural Assessment of the Dysexecutive Syndrome – children's version; CEFA, Cambridge Executive Functioning Assessment; F, F-values, with 1,38 or 2,37 degrees of freedom; r^2, % variance explained.

test, Verbal Fluency, was significantly correlated with BPVS score; and correlations between BADS-C subtests and BPVS scores were similarly unimpressive. Direct comparisons between the present study and that of Ball *et al.* (2008) are possible because both used the CEFA battery. As shown in Fig. 2, the cohort of people with Down syndrome (but without dementia) tested by Ball *et al.* (2008), who had lower receptive language ability than the present mixed cohort, also performed less well on memory tests, but the two groups performed equally well on EF tests, at least in aggregate. This is consistent with the report of Pennington *et al.* (2003) that children with Down syndrome performed less well than a control group matched for mental age on a battery of learning and memory tests, but the Down syndrome group performed at least as well on a battery of EF tests (but with a different pattern of strengths and weaknesses).

Implications of the under-performance of the present sample on the BADS-C

The weakness of the relationship between IQ and EF is further illustrated by small differences in EF across IQ ranges in the normative tables in the BADS-C manual (Emslie *et al.* 2003). The present cohort, with IQs in the 50–70 range, would be predicted to score within the 'average' range relative to the IQ 70–90 cohort. In fact they were severely impaired, equivalent to the lowest 0.1% of the normative sample of people with IQs of 70–90.

The interpretation of this dramatic under-performance is unclear. We were able to rule out three potential explanations, relating to age differences between the present sample and the normative sample, and potential non-linearity of the IQ-EF function. A further consideration is whether the present cohort, consisting of volunteers recruited in day centres, was atypical. However, the normative sample in the BADS-C manual were also volunteers, and there is no reason to suppose that people who attend day centres differ significantly in their cognitive abilities from others known to leaning disability services who do not attend day centres (though they may differ in other respects, such as the extent of support from carers and the prevalence of challenging behaviour).

A more pertinent issue may be that the present cohort was not a random sample of people with IQ < 70, because all participants had a clinical diagnosis of 'learning disability'. Around 2–2.5% of the population have IQs below 70, but only around

0.5% of the population – some 20–25% of people whose IQ is <70 – are known to services. Of those not known to services, some may be unknown because carers choose not to make a referral, but the majority are unknown because they do not meet the clinical criteria for a diagnosis of 'learning disability'. This is because this diagnosis is based not only on a significant impairment of both intellectual ability (defined as IQ < 70), and also functional ability (British Psychological Society 2000), and the majority of the people with IQs below 70 do not meet the latter criterion. To the best of our knowledge, there has been no neuropsychological investigation of the difference between people with IQs of 50–70 who meet criteria for functional impairments and those who do not. (Indeed, very little is known about the basis for this distinction from any perspective, e.g. Zigler 1967.) Nevertheless, the discrepancy in EF between the present cohort of learning disability service users and the predicted performance of a random sample of people in the IQ range 50–70 suggests strongly that poor EF may be an important component of 'functional impairment' (see also Willner *et al.* 2010). As such, it can be hypothesised that EF would be higher in people with IQ < 70 who are not clients of learning disability services, and conversely, that EF might be lower in some groups of 'vulnerable adults' with IQ > 70 who can only function with support from services.

Interpretation of factor scores

There has been considerable debate as to whether EF is a unitary or a multidimensional construct, which is typically addressed by conducting factor analyses of batteries of tests. However, few studies have administered batteries of EF tests to people with IDs, and none have asked this question. The CEFA has only been used in one previous study (Ball *et al.* 2008) and its factor structure has not previously been examined. The adult version of the BADS has been reported to have a unitary structure (Vargas *et al.* 2009) but to the best of our knowledge, the children's version has not been factor analysed. Therefore, the present study may represent the first attempt to examine both the factor structure of EF in people with IDs, and the factor structure, in any population, of the BADS-C and the CEFA.

This is not an ideal dataset for factor analysis because of the relatively high proportions of participants scoring at floor or ceiling on several of the subtests. However, this is almost inevitable when using existing test batteries, and would require extensive further test development to overcome. Fortunately, parametric statistical tests are relatively robust to violations of the assumption of normality (Havlicek & Peterson 1977; Hubbard 1978; Edgell & Noon 1984) and indeed, it is permissible for principal components analysis to include dichotomous variables (where all values are either at 'floor' or 'ceiling') (Kim & Mueller 1978; Comrey & Lee 1992). Nevertheless, the relatively small sample size for this study does mean that the results of these exploratory analyses should be treated as provisional.

Children's version of the Behavioural Assessment of the Dysexecutive Syndrome

The analysis of the BADS-C produced two factors, but one of them was considered invalid, so it is difficult to comment on the structure of this battery. As regards the valid factor, it is not obvious how this should be interpreted, particularly as further analysis showed that it is not robust. The Playing Cards test is a straightforward test of working memory, in which the task is to compare the current card with the previous card. The Water Test is a problem-solving task in which a set of objects (a tube, two lids and a hook) are manipulated to achieve an objective (floating a cork). It is likely that this task also involves working memory when considering alternative strategies. The Zoo Map 1 test is a complex planning task that again engages working memory to organise a sequence of steps. The two planning tasks also involve internal verbal operations and the Playing Cards test requires sustained attention to the instructions. Thus, this factor appears (provisionally: but see below) to reflect working memory as well as language functions, consistent with the strong association between the scores on this factor and receptive language ability (Table 4).

Cambridge Executive Functioning Assessment for people with intellectual disability

The analysis of the CEFA also produced two factors, both of which, in this case, appear to be valid. Factor 1 appears to reflect working memory: the Scrambled Boxes test is primarily a spatial working memory task, and working memory is also a major component of the Tower of London and object memory tasks. The interpretation of Factor 2 is less obvious. However, the involvement of working memory in the Cats and Dogs test is minimal: the test primarily involves behavioural inhibition (saying 'Cat' to a picture of a dog and vice versa), and rule shifting (after the initial phase of naming 'Cat' and 'Dog'); working memory is only used to remember the rule. Similarly, working memory could be considered to be the least important of the several processes involved in Verbal Fluency. Working memory is not involved in Prospective Memory or the Weigl Sorting task. Hence, the common feature of the tasks contributing to Factor 2 may be that they do not significantly involve working memory. It could further be argued that initiative (response initiation or shift) is a common feature of the Factor 2 tests: this is a prominent feature of three of them (Verbal Fluency, Weigl Sorting and Prospective Memory), and also a minor feature of the Cats and Dogs test, given that the first phase of the test involves direct responding ('cat' for cat and 'dog' for dog), with reverse responding introduced subsequently as a rule change.

The two CEFA factors bear an interesting and potentially important relationship to the development of dementia in people with Down syndrome. Ball *et al.* (2008) reported that in people with Down syndrome, those with dementia performed worse than those without dementia on all of the CEFA subtests, but there was considerable variation in the effect sizes. Large effect sizes were seen on the three Factor 1 subtests (range of eta squared values: 0.14–0.20, mean: 0.18), comparable to the decline in performance on immediate and delayed memory tests (range: 0.11–0.27, mean: 0.19). By contrast, only small effect sizes were reported for all four of the Factor 2 subtests (range: 0.04–0.06, mean: 0.05). The observation that EF tasks with a strong working memory component (Factor 1) were more impaired in dementia than EF tasks with a weak working memory component (Factor 2) is consistent with reports that the central executive component of working memory is particularly impaired in Alzheimer-type dementia (Baddeley *et al.* 1986, 1991; MacPherson *et al.* 2007). The differential impairment of these two sets of tasks is also consistent with the observations (Table 4) that Factor 1 (impaired) is associated with delayed memory while Factor 2 (relatively unimpaired) is associated with immediate memory, which is characteristically spared in Alzheimer-type dementia (e.g. Burkart *et al.* 1998; Bayley *et al.* 2000; Foldi *et al.* 2003).

Another difference between the two CEFA factors is the strong visuospatial nature of all of the Factor 1 tests, as distinct from the strongly verbal content of the Factor 2 tests. As such, the Factor 1 tests are likely to involve not only prefrontal cortex, but also temporal cortex, and to be susceptible to the impairment of visuospatial information processing that occurs early in the course of Alzheimer-type dementia (e.g. Gregory & Hodges 1996; Swainson *et al.* 2001; Schmidtke & Olbrich 2007). A perennial problem in this area of research is that the factor structure of the complex set of abilities that constitute EF may depend not only on the cognitive processes that contribute to EF, but also on surface features of the tests, such as the dominant modality in which they are presented and processed (Burgess 1997; Phillips 1997; Miyake *et al.* 2000; Salthouse 2005).

Combined analysis

It had been anticipated that the combined analysis would produce a factor that combined the CEFA Factor 1 with the BADS Factor 2, as working memory was initially considered as the basis for both of these factors. However, while one of the BADS items, Zoo Map 1, did load onto the same factor as the CEFA Factor 1 items, the other two BADS items, Playing Cards and the Water Test did not.

Alternatively, Combined Factor 3 could be considered (following the loss of the Zoo Map 1 test from BADS Factor 2) to represent behavioural inhibition. This is a prominent feature of the Playing Cards test, where, in order to respond to the instruction 'Say yes if this card is the same colour as the previous card (and no if it is not)', the participant must inhibit the response to the earlier rule 'Say "yes" to red and "no" to black'. Inhibition is also a feature of the Water Test, where points are lost for perseverative errors. Viewed in this way, Factors 1 (working memory), 2 (response initiation/

shift) and 3 (behavioural inhibition) resemble quite closely a well-accepted structure of EF, that comprises the three factors of updating (working memory and monitoring), shifting (self-generative behaviour and set shifting) and inhibition of prepotent responses (Miyake *et al.* 2000; Fisk & Sharp 2004; Busch *et al.* 2005b; Collette *et al.* 2005; Verhaeghen *et al.* 2005; Hedden & Yoon, 2006; Friedman *et al.* 2008). The correspondence is not perfect. For example, the Tower of London task loaded on 'updating', whereas Miyake *et al.* (2000) reported that the Tower of Hanoi task (the progenitor of the Tower of London) loaded on the 'inhibition' factor; however, this can probably be explained by a difference in difficulty: the Tower of Hanoi required a minimum of 11 moves to solve versus only 2–5 moves in the Tower of London. Of greater concern, Cats and Dogs, a version of the Stroop (1935) task, might have been expected to load more strongly on 'inhibition' than 'shifting', albeit that the ability to switch between responses is a significant element of this test. Nevertheless, the parallels between the present structure and the conventional model are intriguing, considering that the tests included in this study were selected with no hypothesis about factor structure in mind.

Conclusions

We return finally to the questions posed in the Introduction:

1 People with mild learning disabilities are at the bottom of the viable ability range for the BADS-C, and at the top of the viable ability range for the CEFA. Therefore, the CEFA is likely to be a more useful test battery clinically, as, unlike the BADS-C, it could also be administered to service users in the 'moderate learning disability' range.

2 The extent to which the BADS-C measures a unitary construct of EF remains uncertain, because only one set of three subtests was retained from the initial analysis. However, the CEFA produced two clear factors, which differ in both the extent of involvement of working memory and the predominant sensory modality. The reality of the two factors is supported by their differential impairment in dementia (as seen in the data of Ball *et al.* 2008).

3 BADS Factor 2 and the CEFA Factor 1 were both characterised initially as 'working memory', but in the combined analysis, only one of the three BADS Factor 2 subtests loaded on the same factor as the CEFA Factor 1 subtests. The factor structure revealed by the combined analysis resembles the tripartite structure of EF (updating, shifting, inhibition) proposed by Miyake *et al.* (2000), which has received a broad level of support from studies in the general population.

This study is presented as a first attempt to understand the structure of EF in people with IDs, in the context of the prerequisite need to understand better the two test batteries that we have used. The study has many limitations, including a relatively small sample (for a study of this kind), an ill-defined population of participants (who represent, however, the reality of clinical services for people with IDs), a high proportion of participants with 'floor' or 'ceiling' scores on several subtests, and the exclusion of half of the data from one of the test batteries. The small sample size in particular means that these exploratory results must be considered provisional and in need of replication. Nevertheless, the study has produced results that may be of both clinical and theoretical value, and which provide a basis for future research.

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