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A screening battery for the assessment of executive functioning in young and adult individuals with intellectual disability

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Abstract

Background: According to literature, the assessment of executive functions (EFs) is helpful not only for the diagnosis of brain diseases and of their possible progression over time, but also for describing the ability of an individual to develop and coordinate an adaptive response to the environment. The limited development of EFs has been described in individuals with intellectual disabilities of different ages and aetiology. The aim of the study was to evaluate the usefulness of Frontal Assessment Battery (FAB), a well-known brief battery, for a first assessment of executive functioning in young and adult individuals with intellectual disabilities.
Methods: One hundred and twenty-two young and adult individuals with mild to severe intellectual disability completed the FAB, which included six executive tasks, pertaining to three different domains. Their ages ranged from 18 to 50 years.
Results: FAB scores were significantly decreased in the study participants. The analyses showed that both main domains and specific executive tasks were differently affected in their levels of functioning in terms of age, aetiology and severity of intellectual disability.
Conclusions: The FAB can provide screening information on executive functioning in intellectual disabilities and can help trace dys-executive profiles in adults.

Keywords: assessment; executive functions; intellectual disabilities.

Introduction

Executive functions (EFs) usually refer to cognitive abilities responsible for controlling and coordinating performance in complex cognitive tasks. Goal selection, planning, inhibition of irrelevant impulses and responses, monitoring and regulation of activity, and evaluation of the results of actions are most often included in EFs (1, 2).

The skills subsumed under this term are not homogeneous and have been found to dissociate with focal brain injury (3, 4). The areas involved in their correct functioning are located in the prefrontal cortex and are also characterised by a modular organisation (5, 6). As such, the dys-executive syndrome, which results from damage in these areas, is a complex and heterogeneous condition characterised by several cognitive, behavioural, affective and motivational aspects; here, cognitive components are particularly involved when facing non-routine situations, such as novel, conflicting, or complex tasks (4). According to current theories, the efficient development of each of these components is needed in elaborating appropriate goal-directed behaviours and adapting the subject's response to new or challenging situations.

The role of "executive dysfunction" in developmental disorders, such as autism and attention deficit hyperactivity disorders, has already been demonstrated and these studies have argued the importance of developmentally-appropriate executive capacity even in young children (7, 8). Recent studies on Down syndrome (9, 10) have demonstrated a broad range of executive deficits, while research on Williams' syndrome (11) has shown relatively unimpaired components, i.e., verbal categorisation and shifting. In addition, students with borderline intellectual functioning have shown pervasive executive function deficits (12). Finally, in a recent validation study of two batteries for studying EF in intellectual disability (ID), Willner et al. (13) described a structure of the EF in people with IDs that closely resembles a model of EF in the general population.

This paper addresses the usefulness of utilising a screening battery in assessing the EFs of young and adult individuals with ID and in investigating the role of clinical variables, such as age, degree of disability, and aetiology. A higher level of performance is expected in mild ID; dissociations in functioning should be described in tasks and domains as a function of disease aetiology and severity. Given that EFs are age sensitive (14), the role of age should also be investigated in order to detect changes which, according to typical life-span theories of changes, should be differentially expected.

Frontal Assessment Battery

The Frontal Assessment Battery (FAB) is a brief tool exploring different domains of executive functions. Six subtests explore both cognitive and behavioural domains, each associated with

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specific areas of the frontal lobes on the basis of neuropsychological, electrophysiological, and functional arguments (15). The structure and content of the battery are as follows:

- Conceptualisation: Subjects have to conceptualise the links between two objects from the same category (similarities).
- Mental flexibility: Subjects recall as many words as they can, beginning with a given letter in a 1-min trial (category fluency task).
- Motor programming: Subjects are asked to execute the Luria's motor sequence test consisting of a series of movements in a given correct order (motor series).
- Sensitivity to interference: Subjects are given conflicting instructions and asked to provide an opposite response to the examiner's alternating signal (conflicting instructions).
- Inhibitory control: Subjects must inhibit a response that was previously given to the same stimulus.
- Environmental autonomy: The spontaneous tendency to adhere to the environmental sensory stimulations is analysed (prehension behaviour).

The score for each subtest may vary from 0 to 3, with a score of zero given when the subject fails to provide an answer or responds inappropriately. FAB total score ranges from 0 to 18. The administration of the entire battery requires approximately 10–15 min. The global performance on these six subtests gives a composite score summarising the severity of the dysexecutive syndrome, whereas individual scores might suggest a descriptive pattern of executive dysfunction. In addition, tasks can be grouped into cognitive (conceptualisation and mental flexibility), control (go-no go task and environmental autonomy), and motor programming (motor programming and sensitivity to interference). Thus, the FAB has the potential not only to screen for executive functioning but also to trace a dys-executive profile.

Prior to this use, several authors have examined its properties, including the distribution of scores, internal consistency, relationship to age, and sex. The reliability of the Italian version has also been assessed (16). Inter-rater reliability, which is determined by comparing the scores of two independent raters present during the administration of the FAB by one of them, is optimal. The internal consistency of the battery, i.e., the extent to which the items of the FAB reflect the same underlying construct, is analysed by calculating the Cronbach's coefficient of α, thus showing good internal consistency (α=0.78).

Previous studies have shown that the instrument seems successful in differentiating the frontal dysfunction of patients with cortical and subcortical lesions (17) as well as those with fronto-temporal dementia or supranuclear palsy (18). Studies aimed at identifying the cerebral regions assessed by the FAB have shown that the instrument is an adequate tool for assessing functions specifically related to the dorsolateral and medial frontal cortices and is, therefore, useful in the evaluation of diseases associated with frontal dysfunction (19).

Methods

Participants

The study was carried out with 122 young and adult individuals with ID (56 females and 66 males), as verified by local board-certified local physicians. Mean age for the whole sample was 28.45±7.9 years (age range was from 18 to 50 years). Mean duration of education was 10.8±2.5 years (range 5–13 years). All participants were community-dwelling individuals who were either working in a supported environment or otherwise engaged in occupational programs. Subjects included were at least able to follow simple oral instructions and imitate gestures, as assessed by clinical observations done prior to testing. Raven's Coloured Progressive Matrices (RCPM) (20), for which Italian data were available on intellectual disabilities (21), were used in order to obtain their current global levels of performance. Table 1 summarises the participants' characteristics.

The aetiology of the disability was Down syndrome (DS=42) or developmental ID (i.e., ID affecting the development from the early phases, DID=80). Mann-Whitney U-tests did not show differences between the two groups with respect to Raven's Coloured Progressive Matrices (mean and standard deviation were 12.5±5.99 and 13.06±7.00, respectively). However, the groups significantly differ with respect to age (z=–2.33, p<0.02), which is higher in the DS than in the DID group (26.45±6.89 and 29.51±8.23, respectively). On the basis of the Raven scores, each study participant was allocated to different severity groups defined as mild (Raven score <17; 35 subjects), moderate (Raven score 11–17; 37 subjects), or severe (Raven score <11; 48 subjects).

Data analyses

Kolmogorov-Smirnov Test (KST) was used to estimate the normality of distributions. Non-parametric statistics (χ^2, Mann-Whitney U-test and Spearman's rho) were also used to study the relationship between FAB total score and domains and tasks, age, and degree of disability. SPSS-PC version 16.0 was used for the analyses.

Table 1 Study group characteristics.

Groups	Age, year	Gender		Raven coloured progressive matrices
		Males	Females	
Total group	28.46 (7.91)	66	56	12.52 (6.12)
Down syndrome (DS) (n=41)	26.45 (6.89)	21	21	12.5 (5.99)
Developmental intellectual disability (DID) (n=81)	29.51 (8.23)	45	35	12.53 (6.23)

Means and standard deviations (in parentheses).

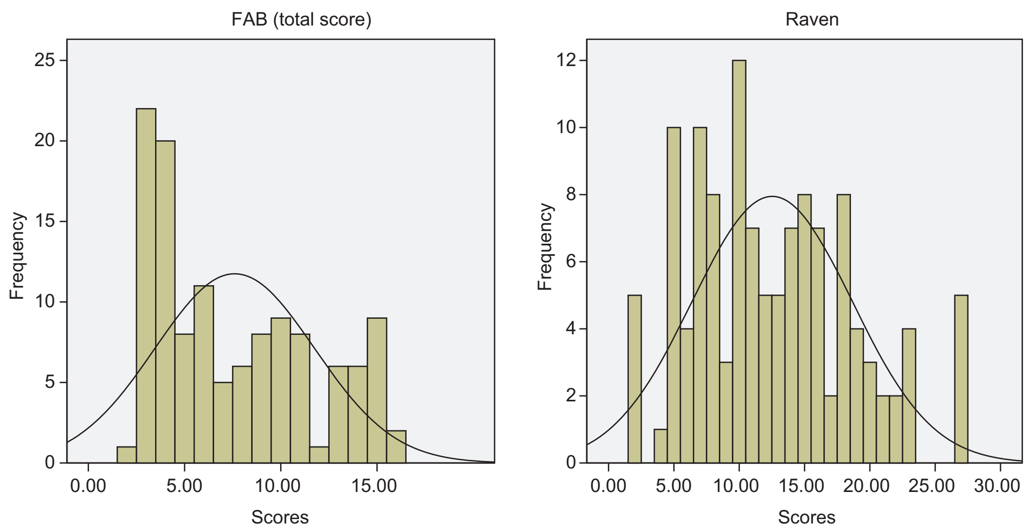

Figure 1 Histograms of the raw score distributions on FAB and RCPMs.

Results

The distribution of the raw scores on FAB and RCPM is shown in Figure 1. Normality assumptions were tested using KST. The distribution significantly departed from normality in the total FAB score, while the normality distribution criteria held for the RCPM (p>0.05). The scores for each of the two disability groups showed a negatively skewed distribution (p<0.01) for the FAB; again this was not found for the RCPM. The accumulation of scores at the lower end was marked, but not restricted to a single value for both instruments. Meanwhile, a bivariate scatter plot (Figure 2) between the FAB and the RCPM scores showed the effect of the degree of intellectual disabilities. As can be seen, the correlation between the two scales was highly significant (r=0.577, p<0.001).

In order to assess the strength of the relationship between the two screening tools, the correlations were also calculated between RCPM and each of the six tasks included in the FAB. Pearson's correlations were highly significant between RCPM and showed respective similarities (r=0.455, p<0.01), fluency (r=0.583, p<0.01) and motor series (r=0.505, p<0.01); they were significant with the go-no-go task (r=0.343, p<0.01) and with the conflicting instructions (r=0.392, p<0.01). A non-significant correlation was found between RCPM and prehension behaviour (r=0.022, n.s.). The analysis of correlations shows a specific relationship pattern between a global cognitive measure, such as RCPM and several executive components. The analysis of the distribution was also carried out for each FAB task (Figure 3).

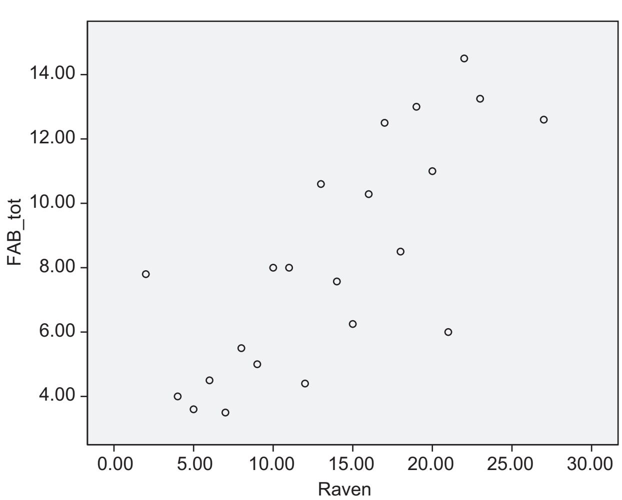

Figure 2 Scatter plots of the FAB and RCPM raw scores.

The accumulation of scores at the lower end is marked for both the a) cognitive and b) control tasks. The distribution was positively skewed for the prehension behaviour, whereas motor series showed a relevant proportion of scores accumulating at both the lower and upper ends of the curve. Grouping tasks according to underlining executive processes (i.e., cognitive, control and motor processes) resulted in Kolmogorov-Smirnov distributions of raw scores obtained in each domain that significantly departed from normality (p<0.01).

Percentage distributions of scores obtained in the six FAB tasks are shown in Table 2. Comparing the results with the distribution of normative data, which was collected for the corresponding age decades by Apollonio et al. (16), only the prehension behaviour task showed an overlapping distribution with the one exhibited by Italian adults with typical development.

In this task, most of the study participants (94.4%) obtained the maximum score and no one reported a score of 0. The lowest percentage of subjects of the normative sample scoring at the optimal level (score = 3) was about 42% for similarities and 65% for fluency. Only 8% showed a 0 score in the most difficult task (similarities), while in the other tasks, the percentage of subjects with a score of 0 was <1%.

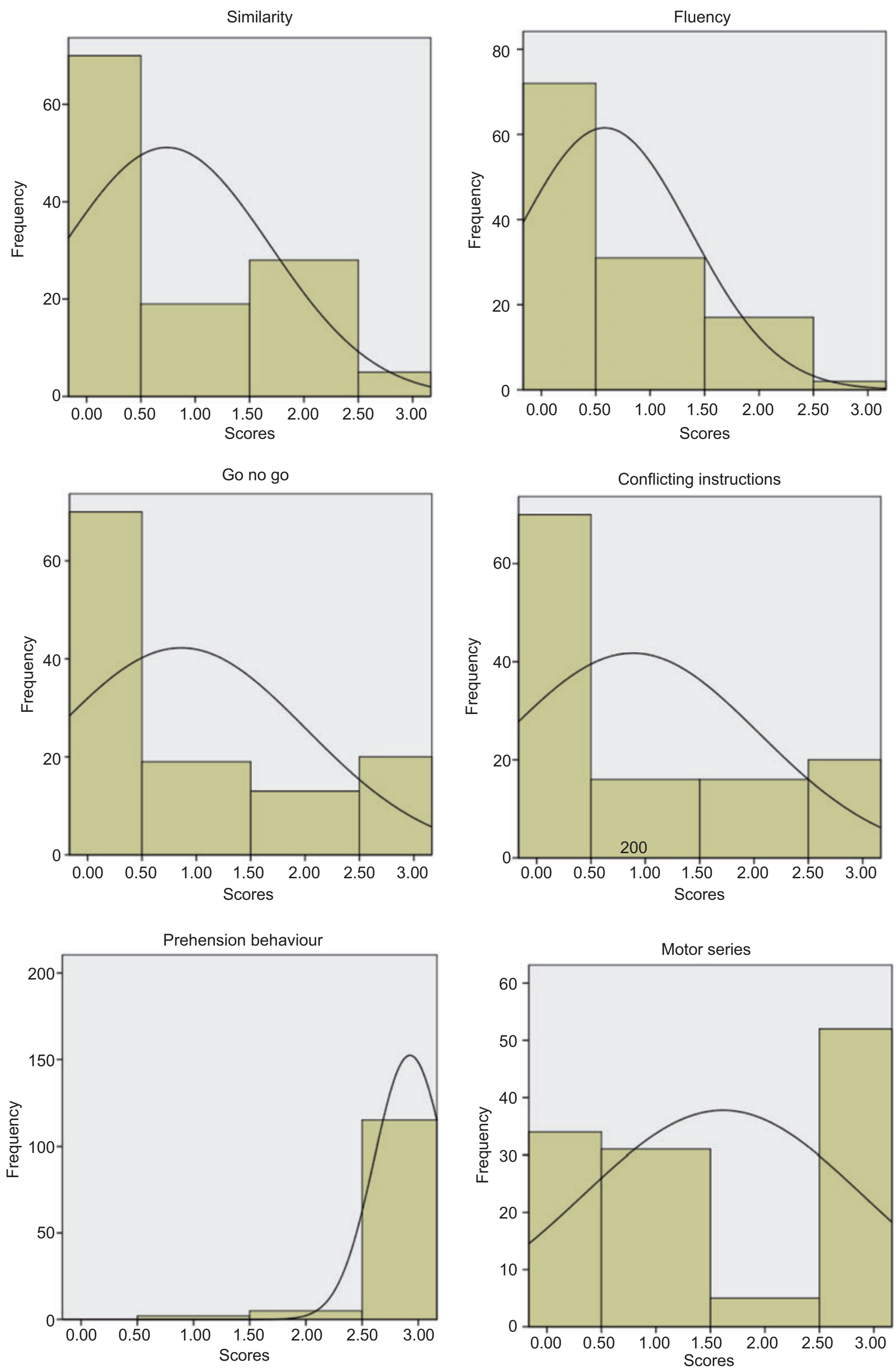

Figure 3 Histograms of the raw score distributions of the subjects on each FAB task.

Executive functions and degree of disability

A non-parametric statistic for n independent samples (Kruskal-Wallis) was used in order to verify differences in FAB scores according to the level of disability. Mann-Whitney test was used to calculate pairwise comparisons, setting the α's level of significance on the basis of the number of comparisons (α=0.05/3=0.01).

Table 2 Percentage distributions of scores obtained by young and adult individuals with IDs.

Raw score	Similarities	Fluency	Conflicting instructions	Go-no go	Prehension behaviour	Motor series
0	57.4	59	57.4	57.4	0.0	27.9
1	15.3	25.4	13.1	15.6	1.6	25.4
2	23.0	13.9	13.1	10.7	4.1	4.1
3	4.1	1.6	16.4	16.4	94.3	42.6

As shown in Table 3, with the exception of prehension behaviour, a general group effect was detected for each executive task. Mean scores obtained by subjects at different levels of disability suggested a progressive decrease in performance, corresponding to an increase in the disability level. In order to further analyse differences between groups, Mann-Whitney comparisons were again conducted on single FAB tasks. Group effect was found for all tasks, except for prehension behaviour. In addition, significant differences were found in pairwise comparisons between the three groups on cognitive and motor domains ($p<0.01$). Control showed a significant effect ($p<0.01$) when comparing mild to severe groups and moderate to severe groups, but not when comparing mild to moderate groups (Table 4). Finally, when looking at motor domain, motor programming was mainly responsible for the differences evidenced across levels of disability; in fact, no differences were found across level of disability in pairwise comparisons conducted on scores obtained in prehension behaviour task.

The role of aetiology In order to verify the possible role of aetiology, non-parametric analyses for independent samples were conducted on FAB total scores and on subtests comparing both Down syndrome condition and DID (see Table 5). Mann-Whitney comparisons did not show significant differences on specific executive tasks, except for motor series where a higher mean score was evidenced in DS ($z=-2.01$, $p<0.04$). No differences were found when comparing executive domains.

Gender and age effects Gender effect was analysed on the total sample (Figure 4). Mann-Whitney comparisons did not evidence significant differences according to gender, both on single tasks and on executive domains. Subjects were then divided into three groups in order to analyse possible age changes. These groups were also comparable with respect to cognitive level, as shown by the Mann-Whitney test results. Meanwhile, KST results evidenced a general age-group effect only on similarities ($\chi^2=9.95$; $p<0.01$) and on motor programming score ($\chi^2=10.04$; $p<0.01$).

Mann-Whitney post hoc comparisons showed significant differences when comparing the youngest (18–25 years old) and the intermediate age group (26–35 years old) in terms

Table 3 Means and standard deviations (in parentheses) of FAB total score of the sample according to levels of ID on broader FAB domains.

Domains	Degree of disability			Group effect	Mild-moderate	Mild-severe	Moderate-severe
	Mild (n=28)	Moderate (n=41)	Severe (n=53)				
Cognitive	2.78 (1.59)	1.34 (1.44)	0.53 (1.14)	39.43[a]	–3.56[a]	–6.11[a]	–3.39[a]
Control	4.21 (1.61)	3.95 (1.18)	3.43 (0.89)	7.38[b]	–0.96 n.s.	–2.40[b]	–2.07[b]
Motor programming	4.07 (1.80)	2.78 (1.94)	1.45 (1.66)	30.89[a]	–2.61[a]	–5.24[a]	–3.39[a]

[a]p<0.01, [b]p<0.05. n.s., not significant.

Table 4 Means and standard deviations (in parentheses) of FAB scores, according to levels of ID on the six FAB tasks.

Executive tasks	Degree of disability			Group effect	Mild-moderate	Mild-severe	Moderate-severe
	Mild (n=28)	Moderate (n=41)	Severe (n=53)				
Similarities	1.46 (0.96)	0.76 (0.89)	0.34 (0.76)	27.30[a]	–2.95[a]	–5.01[a]	–2.81[a]
Fluency	1.32 (0.86)	0.59 (0.71)	0.19 (0.48)	37.77[a]	–3.47[a]	–6.00[a]	–3.27[a]
Conflicting	1.64 (1.28)	0.93 (1.17)	0.45 (0.87)	17.53[a]	–2.22[a]	–4.14[a]	–2.15[b]
Control	1.36 (1.39)	0.98 (1.17)	0.51 (0.87)	7.31[a]	–1.17 n.s	–2.51[a]	–1.80 n.s
Motor series	2.42 (0.92)	1.85 (1.3)	1.0 (1.16)	25.23[a]	–1.99[b]	–4.87[a]	3.04[a]
Prehension	2.86 (0.52)	2.98 (0.16)	2.92 (0.27)	1.26 n.s.	–0.96 n.s.	–0.22 n.s	–1.08 n.s.

n.s., not significant.

Table 5 Means and standard deviations (in parentheses) of FAB scores obtained by the two groups in each task.

Executive tasks	Down syndrome (n=42)	DID (n=80)	Group differences
Similarities	0.83 (0.91)	0.68 (0.97)	n.s
Fluency	0.48 (0.67)	0.63 (0.84)	n.s
Conflicting instructions	0.67 (0.90)	1.00 (1.27)	n.s
Control	0.67 (0.90)	0.80 (1.19)	n.s
Motor	1.95 (1.23)	1.29 (1.29)	[a]
Prehension	2.90 (0.3)	2.93 (0.33)	n.s

[a]p<0.01. n.s., not significant.

of motor programming (z=–2.85; p<0.01), with the younger group scoring at a higher level. No differences emerged when comparing the intermediate and oldest subjects (36–45 years old). Finally, when comparing the youngest and oldest subjects (18–25 and 36–50) significant differences in terms of similarities (z=–3.00; p<0.001) and motor programming score (z=–2.40; p<0.01) emerged once again.

Discussion

The FAB is easy to administer, and the components tapped by the battery were particularly involved in any atypical situation, such as novel, conflicting, or complex tasks and relevant when performing many complex daily tasks. The FAB was designed for use by clinicians and was intended for serial use in order to detect cognitive decline – also a relevant issue for young and adult individuals with IDs (22–24). It differs from standard tests of intelligence in that most of the items seem to be sensitive to oncoming damage. In comparison with neuropsychological batteries, the FAB is brief and is capable of minimising the effect of wandering attention or fatigue.

In our sample of young and adult individuals with IDs, reduced performance was evidenced in five out of the six tasks. With respect to cognitive domain, both tasks showed a low level of performance: adults with ID had limited abilities in establishing an abstract link between the items, finding a

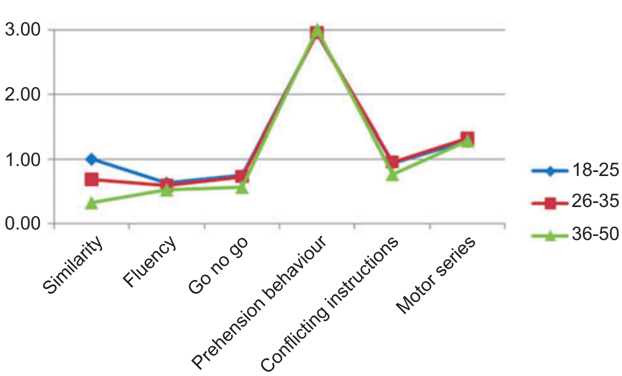

Figure 4 Mean scores according to age groups.

link of similarity (category fluency), and dealing with non-routine situations, in which self-organised cognitive strategies had to be identified and strengthened (mental flexibility). Deficits in behavioural self-regulation were also evidenced by the difficulties met in inhibiting the natural tendency to repeat the same movement performed by the examiner and – similar to patients with acquired lesions in the frontal lobe – in inhibiting inappropriate responses.

With respect to motor domain, two different patterns emerged. Frontal lobe damage may impair tasks requiring temporal organisation, maintenance and execution of a series of actions: this seemed true for subjects with moderate to severe disability. Meanwhile, the performance of subjects with mild ID and adults with Down syndrome in the motor series task was close to that of normal controls. Patients with specific frontal lobe damage were found to be dependent on environmental cues; as a consequence, they showed a spontaneous tendency to adhere to the environment sensory stimulations. Nevertheless, this type of deficit was not found in almost all study participants, showing a robust environmental autonomy.

As expected, RCPM strongly correlated with the FAB scores since both tests were designed to measure cognitive functioning. The highly significant correlations found for both similarities, namely, fluency and motor series, showed that RCPM and FAB activated and shared some, but not all, common executive resources in accomplishing the task.

One of the aims of the present study was to examine the effect of general ID on test performance. The level of disability into which the cohort was divided, based on RCPM, was the best estimate of the degree of pre-existing disability available to the authors, and was completely independent of the study programme. Changes detected according to the degree of disability were evidenced with the exception of prehension behaviour task (where the performance was stable across groups identified) and in the control task (where differences where found only when compared with mild to severe ID).

Our results also showed that there was a greater difficulty in developmental ID than in the Down syndrome group, and it seemed to be determined by a more limited efficiency of motor performance. It is possible that more differences may have been masked by the heterogeneity of the DID group. Moreover, the cognitive components of the executive skills investigated by FAB seemed more sensitive to age changes. This result is particularly relevant because the two groups showed a comparable ID severity. These are processes that should be monitored in order to find information about cognitive decline expected with increasing age. The result is in agreement with the international guidelines for the diagnosis of dementia, which requires the systematic and formal evaluation of EFs in the work-up of any given subject with a clinical suspicion of progressive decline (17–19).

In summary, FAB seems a useful instrument in the assessment of executive functions. It can describe executive skills in adults with developmental ID, from whom marked deficits and well-developed abilities can be observed. Both domains investigated and specific tasks included in the battery seemed to be differently affected by age and severity of ID. The study underlines the need to evaluate different aspects of cognition to identify specific strengths and weaknesses that may underlie adaptive behaviour dysfunction.

Evidence from literature seems to suggest the relevance of systematically assessing executive functioning and the appropriateness of following general guidelines used in neuropsychological assessment, which includes both screening and subsequent in-depth testing. In order to fully understand the ability of a person with ID to function effectively in everyday environment, an executive screening based on FAB should be followed by an extended assessment, possibly conducted using a naturalistic battery (25). A longitudinal follow-up using FAB can certainly contribute to evidence subtle manifestations of preclinical or incipient dementia.

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