Chapter 4

Effects of a multifaceted treatment program for Executive Dysfunction after Acquired Brain Injury on indications of executive functioning in daily life

Introduction

Executive dysfunction (ED) is a frequent and disabling consequence of acquired brain injury (ABI), impairing patients in their abilities to function independently in daily life.

Executive functions (EF) are those capacities that make persons effective in the real world, allowing them to adapt to new situations and to develop and follow their life goals in a constructive and productive way (Burgess & Simons, 2005b). EF is in fact an umbrella term which encompasses a broad range of higher order capacities for planning, initiation, regulation and verification of complex, goal directed behaviour (Lezak, 1982). These executive capacities are operational in situations that are new, complex and can not be dealt with in a routine matter, in other words, unstructured situations that do not provide unequivocal cues guiding the subject's behaviour. EF are subserved by prefrontally driven brain systems in which other cerebral and cerebellar areas take part (Lichter & Cummings, 2001; Sbordone, 2000). It follows that ED does not only have to be the consequence of injuries directly affecting the prefrontal cortex, but can also result from lesions elsewhere in the brain. Hence, ED can be found in different categories of brain injury patients irrespective of aetiology or lesion location. ED has been extensively documented in TBI patients (Bamdad, Ryan, & Warden, 2003; Bennett et al., 2005a; Busch, McBride, Curtiss, & Vanderploeg, 2005; Hart, Whyte, Kim, & Vaccaro, 2005) with evidence for even more severe problems in case the patients had also focal frontal damage (Fontaine et al., 1999; Spikman et al., 2000a). Ample evidence of ED has also been found in patients with brain injury due to other aetiologies, for example stroke (Leskela et al., 1999; Pohjasvaraa et al., 2002; Sachdev et al., 2004), cerebral tumours (Goldstein, Obrzut, John, Ledakis, & Armstrong, 2004; Tucha, Smely, Preier, & Lange, 2000) and post-anoxic encephalopathy (Armengol, 2000; Simo-Guerrero et al., 2004).

Many ABI patients are referred for rehabilitation to learn to deal with the consequences of their brain injury. Intact EF are crucial: by their very nature they are the capabilities that enable subjects to change and adapt behaviour to altered situations. Hence, EF are decisive in whether impairments result into disabilities or handicaps. In terms of the WHO ICF framework (World Health Organization, 2001), ED does not only affect functioning on activity level, referring to the execution of goals in tasks or actions, but also on participation level, that is patient's fulfilment of social and vocational roles in everyday life. ED is negatively related to indications of activity and social participation level, for example occupational functioning, psychosocial adjustment, community integration and coping abilities (Anson & Ponsford, 2006; Crepeau & Scherzer, 1993; Eriksson, Tham, & Borg, 2006; Krpan, Levine, Stuss, & Dawson, 2007; Marsh & Martinovich, 2006; Ownsworth & Fleming, 2005; Ponsford et al., 2008; Reid-Arndt, Nehl, & Hinkebein, 2007; Vilkki et al., 1994; Wells, Dywan, & Dumas, 2005).

The main goal of rehabilitation practice is teaching patients independent living skills. ED constitutes a major obstacle to learning, and subsequently to successful community re-entry (Fasotti & Spikman, 2002). Therefore, effective interventions aimed at improving EF in daily life are sorely needed. However, difficulties with learning and applying training principles are inherent to ED. Hence, designing clinically relevant interventions requires accounting for those factors that make EF such complex and at the same time essential functions.

The first factor is the heterogeneous construct of EF, encompassing a range of different subfunctions. Ylvisaker (Ylvisaker, 1998) distinguishes the following EF aspects: *self-awareness* of strengths and needs, realistic and concrete *goal-setting*, *planning* the steps to these goals, *self-initiating* these plans, *self-monitoring* and evaluating performance according to plan and goal, *self-inhibiting* behaviour not leading to the goals set, *flexibility and problem solving* when situations can not be dealt with according to plan, and finally, *strategic behaviour*, transfer of successful behaviours to other situations. These aspects can be differentially impaired, leading to different patterns of EF symptoms in patients. Consequently, clinically relevant treatments should be *multifaceted* and aimed at improving a comprehensive but finite range of EF. So far, such evidence-based protocols are sparse (Cicerone et al., 2005; Cicerone et al., 2000). Only a few studies have been carried out addressing a limited set of EF aspects, like problemsolving (Cramon von et al., 1994; Foxx et al., 1989), goal management (Levine et al., 2000) or self-regulation (Medd et al., 2000).

Another point is the level of functioning on which interventions target. Interventions should be ecologically valid and have impact on behaviour in the real world (Worthington, 2005). This usually involves teaching compensatory cognitive strategies; top-down approaches, which have to be adapted and applied to the different problems that patients encounter (Fasotti et al., 2002). However, common EF deficits hampering *transfer* to the home situation (Geusgens, Winkens, Heugten van, Jolles, & Heuvel van den, 2007) are a lack of *self-awareness* and a difficulty to *self-initiate* behaviour (Fischer et al., 2004; Hart et al., 2004; Marin, 1997; Prigatano, 1991). Therefore, treatment of deficits in self-awareness and self-initiation should be an integral element of any intervention for ED aiming at improvement of daily life executive functioning.

A third factor concerns the measurement of effects of interventions targeted at a broad range of executive aspects. Conventional neuropsychological tests tapping single executive aspects are not likely to uncover these effects. Even more so, the ecological validity of many EF tests (the WCST, Strooptest or Trailmakingtest) remains poor, as these tests do not tap the abilities for self-initiation and self-structurization required in daily life (Burgess et al., 2006a; Burgess et al., 1998; Mountain & Snow, 1993; Spooner & Pachana, 2006). The BADS (Wilson et al., 1996) was specifically designed to be ecologically valid, but evidence for this validity is still inconclusive (Norris et al., 2000a; Wood et al., 2006). A more general problem with neuropsychological tests as outcome measures are test-retest effects of unknown magnitude that may complicate measurement of treatment effects. Hence, such tests are not always appropriate for measuring effects of ecologically valid interventions. This should not be problematic, as the main aim is not that patients improve their test performances, but their daily life skills. Consequently, treatment effects should be measured in terms of improvement on indications of daily life functioning, which are neglected outcome measures (Cicerone,(Cicerone, 2004).

In this paper, a newly developed multifaceted treatment of ED will be presented and evaluated. Herein ABI patients are trained to cope with problems in all eight EF aspects distinguished by Ylvisaker, including diminished self-awareness and lack of initiative, with the final aim to improve everyday executive functioning. This training was given to ABI patients who were expected or were already able to resume (part of) their previous daily life activities. The effectiveness of this multifaceted ED treatment was investigated in a multicenter RCT. Multifaceted training was compared to a control intervention, consisting of a computerized training (Marker, 1987) aimed at improving general cognitive functioning. Since there is no substantial evidence (Cicerone et al., 2000; Cicerone, 2004) that such training programs improve basic cognitive capacities, no effects of this control training were expected.

In order to measure treatment effects comprehensively, we chose our primary outcome-measures as follows. Three measures assessed daily life executive functioning. As these outcome measures covered different aspects of executive functioning on both activity and participation levels, we considered them as complementary, which allowed us to combine them into a single, main outcome measure.

Our hypothesis was that multifaceted executive training would significantly improve executive functioning in daily life activities and increase social participation. Both effects were supposed to be present immediately after training. However, the treatment aim was to teach patients to apply compensatory strategies to their daily life activities independently. Hence, we considered the effects in the long term as an indication of this ability and expected them to still be present at follow up.

Methods

Study Design and procedure

This study employed a prospective multicenter RCT with two groups of patients receiving treatment, taking place in eight rehabilitation or academic centres in the Netherlands. Data were obtained according to the ethical regulations of these institutions, in compliance with the Helsinki Declaration. Participants eligible for the study had to suffer from Acquired Brain Injury (ABI) of non-progressive nature (i.e. TBI, Stroke or cerebral tumours), with a minimal time post onset of three months and no maximum, their age range had to be 17-70 and they had to live at home. Candidates should be referred for outpatient rehabilitation treatment because of post-injury problems of a clearly dysexecutive nature, either reported by themselves or observed by others, hampering resumption of previous activities and roles. If patients gave their informed consent to participate they subsequently underwent a neuropsychological examination to determine whether final inclusion could take place.

Dysexecutive problems were measured by means of the Dysexecutive Questionnaire (DEX; (Burgess et al., 1996). Final inclusion was based on the following criteria: a BADS standard age score below average, or a discrepancy between BADS standard age score and IQ (a shortened version of the Groninger Intelligence Test(Luteijn & Ploeg van der, 1983)) of 15 points (1 sd) , or standard scores of 2 or lower on both SET and Zoo Map.

Exclusion criteria were: cognitive comorbidity interfering with the treatment, severe psychiatric problems, neurodegenerative disorders and substance abuse.

Excluded patients were offered standard rehabilitation treatment. When included, patients were per centre blindly and randomly assigned to either the experimental or the control condition.

In each treatment condition, patients underwent 20-24 one-hour treatment sessions, twice a week, during a three month period. At baseline (T0), immediately post treatment (T1) and follow-up six months post treatment (T2), an extensive test battery was administered. During the follow-up period patients underwent no other treatments.

Patients

Seventy-five patients were included, underwent the treatment and were assessed after training. Thirty-eight patients received the experimental treatment and thirty-seven the control treatment, Cogpack (Marker, 1987). Table 1 shows the characteristics of both patient groups. Although there are small differences between the groups, statistical testing (Mann-whitney U and Chisquare tests) revealed these were not significant, which is taken to indicate that both groups were matched well enough. At follow-up, three patients in the experimental group did not return because they did not have the energy to undergo a final, strenuous, assessment and one control patient due to logistical problems in the rehabilitation centre.

Table 1 *Demographic data of the control patient group and experimental patient group.*

	Control patients (n=37)	**Experimental patients (n=38)**	**Significance**
Age *(*M, *sd,* range*)*	43.7 (*14.9*, 17-64)	41.4 (*12.1,* 17-65)	n.s.
Education *(*M, *sd,* range*)*	4.8 (*1.2*, 2-7)	5.2 (*1.0,* 3-7)	n.s.
M/F (%)	65/35	68/32	n.s.
Chronicity (months) *(*M, *sd,* range*)*	47.9 (*64.1*,4-288)	71 (*105.4*,3-468)	n.s.
Etiology (%) (TBI/stroke/other)	32.5/54/13.5	55/32/13	n.s.

Experimental Treatment

The Multifaceted Treatment of Executive Dysfunction's main objective is that patients learn to cope with planning and regulation problems with the final aim to improve everyday executive functioning. The treatment is multifaceted, in that it is directed at improvement of all eight aspects of EF defined by Ylvisaker (Ylvisaker, 1998). The protocol is comprised of three modules, namely 1) Information and Awareness, 2) Goal Setting and Planning, and 3) Initiation, Execution and Regulation. The content of the treatment and modules is described in Appendix 1.

Control Treatment

The control treatment was Cogpack (Marker, 1987), a computerized cognitive training package consisting of several repetitive exercises. It is aimed at improving general cognitive functioning (like reaction speed, attentional functioning, memory and planning). The program is self-supporting and patients can perform the tasks without external help. Task performance is followed by direct feedback from the computer program so that improvements over time can be monitored, but no clues about strategic approaches to the proposed tasks are offered.

Measures

An extensive battery of tests and questionnaires was administered to the patients (see also Boelen, Spikman, Rietveld, & Fasotti, 2008). For this study, the following were relevant.

Primary outcome measures

In order to measure executive functioning on participation level, **The Role Resumption List** (Spikman et al., 2003) was administered at all three testmoments. Based on a structured interview it is rated on a five-point scale (0= no change, 4= severe loss of independence) whether there have been changes for four daily life domains (vocational functioning, (Zomeren van & Burg van den, 1985)), leisure activities, social interaction with partner and family, and mobility) in amount and quality of activities compared to premorbid levels. The scores are added up to a total score with a range of 0 to 16.

Another important measure for the evaluation of the treatment concerned a variant of the **Goal Attainment Scale** (GAS), (Rockwood, Joyce, & Stoleee, 1997). Halfway during both treatments, each patient was asked to determine three personal goals he/she wanted to accomplish by means of the training. At post-measurement and follow-up, patients filled in on a 5-point scale (1= not at all, 5= entirely) to which extent they had attained each of the three goals; these were added up to a total score with a range of 3 -15.

In order to measure EF in a complex task, a newly designed test was administered at folllow-up only. The **Executive Secretarial Task** (Spikman et al., 2007) is comparable to the Multiple Errands Tasks (Shallice et al., 1991) or the Hotel Task (Manly et al., 2002). It requires the organization and prioritisation of multiple tasks over a longer time span than usual, while dealing with delayed intentions, interruptions and deadlines. The task yields a Totalscore as well as three subscores: (*Initiative*, *Prospective*; *Executive)*. The intention was to obtain a "pure" indication of executive functioning at follow-up, without contamination of earlier test knowledge or experience. The test is extensively described in appendix 2.

Secondary outcome measures

Questionnaires and observation lists The presence of executive symptoms in everyday life was investigated by means of the **Dysexecutive Questionnaire** (Burgess et al., 1996), with a patient, proxy and therapist version.

The **Executive Observation Scale** (Pollens, McBratnie, & Burton, 1988)) consists of 8 items covering the EF aspects that Ylvisaker distinguished. Items are rated by a therapist on a scale from 1 (complete inability) to 4 (complete independence) resulting in a total score ranging from 8 to 32.

Quality of life was measured with the **QOLIBRI** (Quality of Life after Brain Injury (VonSteinbüchel, Petersen, & Bullinger, 2005)), consisting of two parts; a satisfactionscale (higher score indicates more satisfaction) and a burdenscale (higher score indicates higher burden).

Only at follow-up, patients were requested to rate the levels of satisfaction about their treatment (results) on a 5-point **Treatment Satisfaction Scale (TSS),** ranging from score 1 (not satisfied) to score 5 (very satisfied).

Neuropsychological measures of executive and cognitive functioning In addition to the behavioural measures, the following EF tests were administered. The **Behavioural Assessment of the Dysexecutive Syndrome** (Wilson et al., 1996); all six subtests, resulting in an Standard Age Profilescore. The **Trail Making Test** (ratio B versus A, **TMT B/A**), **Stroop Test** (Stroop, 1935) (ratio time part three versus time part two (**STR III/II**); **Tower of London** (Shallice, 1982), (**number correct**).

To control for possible effects of the CogPack training on memory, the **15 Words Test** (Dutch version RAVLT (Deelman et al., 1980); immediate (**Memory IR)** and delayed recall score (**Memory DR**)), was administered at baseline and T1.

Statistical analyses

T tests were used to compare patients' scores on relevant measures at baseline with those of healthy controls, as well as to control whether both patient groups differed at baseline.

Treatment effects were analyzed as follows. The primary outcome measure (Total Executive Outcome Score, TEOS) was a sumscore, composed of the standardized values on the EST, GAS and the RRL at follow-up. Before standardizing the RRL, scores were recoded, so that a high score indicated a good performance. A *t* test was applied in order to test whether both groups differed on this sum score. Also an effect size (Cohen's d) was calculated using the difference between the mean scores of both groups divided by the pooled standard deviation. According to Cohen an effect of 0.2 is small, 0.5 is medium and 0.8 is large ((Cohen, 1988).

The secondary outcome measures, as well the results on the RRL separately, were analyzed using repeated measures analyses (GLM repeated measures, SPSS 12.0). Because data loss due to missing values was undesirable, all test-measures were analyzed separately in a univariate design. We performed three series of analyses. First, it was tested whether the scores at T1 were different from baseline, visible in an effect of time, and whether there was a difference in improvement over time between the experimental patients and the control patients, reflected in an interaction effect. Similarly, scores at T2 were compared to baseline performance, as well as to performance on T1. Because there were no baseline measures for the GAS and the EST, results of both patient groups at T1 and T2 were compared using *t* tests. With respect to the EST, results of both patient groups were also compared with those of a group of healthy controls. Finally, the scores on the satisfaction list, being skewed, were compared using a non-parametric method, the Mann-Whitney U test.

Results

In order to check whether the patients had indeed impaired EF and thus fulfilled the inclusion criteria, their BADS and DEX scores were compared to those of a group of healthy controls.

Table 2*Means (and standard deviations) of patients and healthy controls on demographic variables (age, education) as well as on a test (BADS) and questionnaire (DEX) for executive functioning. Two-tailed results of t tests or Mann-Whitney U tests.*

	Healthy Controls (n=57) *(M, sd)*	**Patients (n=75)** *(M, sd)*	**Significance**
Age	47.8 (*11.4*)	42.5 (*13.6*)	p<.05
Education (2-7)	5.2 (*1.0*)	5.0 (*1.1*)	n.s.
BADS Age score	102.3 *(12.3)*	88.3 *(13.0)*	p<.001
DEX patient	18.3 *(8.6)*	31.6 *(13.3)*	p<.001
DEX proxy	18.1 *(9.9)*	32.0 *(14.8)*	p<.001
DEX therapist	10.1 *(6.5)*	35.3 *(12.1)*	p<.001

* p< .05 ** P< .01 ***p<.001

Table 2 shows that both groups were comparable with respect to educational level, but that the healthy control group was slightly older. However, as age is known to influence executive functioning negatively, being older was not advantageous for the healthy controls. With respect to executive functioning, the patients had indeed significantly lower scores on the BADS, indicating worse executive functioning than healthy controls. At the same time they had higher scores on the three DEX measures evidencing more executive problems in daily life.

After inclusion, patients were randomly assigned to one of both treatment conditions. Table 3 shows that at baseline no significant differences were found between the experimental and control patient groups on several relevant measures, such as indications of cognitive functions (IQ, memory) and EF (Stroop 3/2, Trails B/A, Tower of London, BADS). Neither were there significant differences with respect to questionnaires measuring dysexecutive complaints (DEX patient, proxy and therapist), executive functioning observed during task performance by therapists (EOBS), the extent to which previous roles were resumed (RRL) and the satisfaction part of the Quality of Life questionnaire (Qolibri). Only with respect to the Qolibri Burden score a significant difference was found, which indicated that before treatment the experimental patients experienced a higher burden due to their brain injury than the control patients.

Table 3 *Means and standard deviations of test-sores for the control and the patient group at baseline (T0). Two-tailed results of t tests or Mann-Whitney U tests.*

	Control patients (n=37) *(M, sd)*	**Experimental patients (n=38)** *(M, sd)*	**Significance**
Shortened IQ	109 *(14.4)*	116.1 *(16.8)*	n.s.
Memory IR	38.6 *(10.1)*	39.0 *(11.5)*	n.s.
Memory DR	7.9 *(3.6)*	7.6 *(3.3)*	n.s.
Stroop 3/2	1.7 *(0.3)*	1.6 *(0.3)*	n.s.
TMT-B/A	2.3 *(1.1)*	2.1 *(0.6)*	n.s.
TOL nr correct	10.8 *(1.1)*	10.6 *(1.5)*	n.s.
BADS	85.9 *(14.3)*	90.6 *(11.4)*	n.s.
DEX patient	31.0 *(13.7)*	32.2 *(13.1)*	n.s.
DEX proxy	32.1 *(15.5)*	32.1 *(14.3)*	n.s.
DEX therapist	35.7 *(11.7)*	34.9 *(13.3)*	n.s.
Qolibri sat	124.7 *(2,9)*	120.4 *(27.7)*	n.s.
Qolibri burden	48.1 *(13.1)*	54.9 *(11.5)*	**
EOBS	20.6 *(3.9)*	20.7 *(3.2)*	n.s.
RRL	7.8 *(3.6)*	7.2 *(3.0)*	n.s.

* p< .05 ** p< .01 ***p<.001

Effect of treatment

Our primary outcome measure (TEOS) was composed of the results at follow-up (T2) on three effect measures (EST, GAS and RRL) pertaining to different aspects of executive functioning. Table 4 shows that the experimental patients score significantly higher on this measure than the control patients. According to Cohen's criteria the effect size is large, suggesting a substantial effect of the experimental treatment.

Table 4 *Results of t test on the difference between de control patients (CP) and experimental patients (EP) on the TEOS as well as the effectsize for this difference.*

TEOS	**CP**	**EP**	**T**	**Sign.**	**Effect Size**
	-.91 (2.1)	.92 *(1.7)*	3.9	.000	0.87

Table 5 *Means, standard deviations and difference scores (T1-T0) of the test-variables at T1 for the control and experimental patient group. Repeated measures analyses on the test-scores at T1 compared to T0. Two-tailed result of t test on the GAS score.*

	Control patients (n=37)		**Experimental patients (n=38)**		**ANOVA**	
					Time	**Time X group**
	M, *sd*	**M T1-T0**	**M, *sd***	**M T1-T0**		
Memory IR	43.1 *(11.4)*	4.5	45.1 *(11.5)*	6.1	*	n.s.
Memory DR	8.0 *(3.8)*	0.1	8.7 *(3.7)*	1.1	n.s.	*
Stroop 3/2	1.6 *(0.3)*	0.1	1.5 *(0.2)*	-0.1	n.s.	n.s.
TMT- B/A	2.2 *(0.9)*	-0.1	1.9 *(0.7)*	-0.2	n.s.	n.s.
TOL nr correct	11.3 *(0.9)*	0.5	10.9 *(1.6)*	0.2	n.s.	n.s.
BADS	93.8 *(17.5)*	7.9	100.6 *(13.0)*	10.0	***	n.s.
DEX patient	27.2 *(14.6)*	-4.4	26.3 *(15.9)*	-4.8	***	n.s.
DEX proxy	29.3 *(16.5)*	-3.0	26.8 *(16.9)*	-4.0	**	n.s.
DEX therapist	33.3 *(12.2)*	-2.8	26.3 *(12.1)*	-8.6	***	**
Qolibri satisfac	133.4 *(29.4)*	6.6	132.7 *(32.4)*	6.0	**	n.s.
Qolibri burden	45.7 *(10.6)*	-3.7	49.3 *(13.9)*	-6.0	**	n.s.
EOBS	21.2 *(3.7)*	0.6	24.7 *(4.0)*	4.0	***	***
RRL	7.2 *(3.3)*	-0.1	6.4 *(3.2)*	-1.3	**	**
GAS	8.2 *(1.8)*		10.4 *(2.5)*		T = 4,4	***

* p< .05 ** p< .01 ***p<.001

Table 5 shows the results of the repeated measures analyses on the several measures at T1 compared to baseline (T0). On the standard EF tests (Stroop, Trailmaking Test and Tower of London), neither time nor interaction effects were found. As to the BADS, a large improvement over time was found for both groups, but the experimental patients had not improved significantly more than the control patients. On the indications for daily life executive functioning, significant effects were found. Both groups had resumed their previous roles as measured with the RRL significantly more after treatment, but the experimental patients did this to an even larger extent. Also, the experimental patients had attained their goals set early in the treatment to a significantly larger extent than the control patients (GAS). Furthermore, both the DEX patient and the DEX proxy showed a similar decrease of executive complaints for both groups. The DEX therapist revealed that both groups had less executive problems, but the decrease of executive problems was significantly larger in the experimental patients. With respect to quality of life, no effects were found on the Qolibri Satisfaction scale. However, on the Qolibri Burden scale both groups reported the same reduction of burden after treatment. On the Executive Observation Scale (EOBS), a significant effect of time indicates that the executive abilities of both groups, as observed by professionals, had improved, whereby the interaction effect indicates that this improvement was significantly larger in the experimental patients. The Memory IRscore showed a similar improvement over time for both groups which was interpreted as a retest effect. Surprisingly, only the experimental patients improved significantly over time on the Memory DRscore.

Table 6 shows the results of repeated measures analyses at T2 compared to T0. With respect to the neuropsychological EF tests including the BADS, results were similar to those found at T1. This was also the case for the DEX patient and proxy. However, the group difference on the DEX therapist had disappeared at T2. Regarding the Qolibri, both groups reported to experience the same increase in satisfaction when compared with baseline, but the burden scale showed no differences between the groups. Similar to T1 both groups showed improvement on the EOBS and the RRL and on both measures the experimental patients had improved significantly more than the control patients. Again, the experimental patients had a significantly higher score on the GAS than the control patients.

Table 6 *Means, standard deviations and difference scores (T2-T0) of the test-variables at T2 for the control and experimental patientgroup. Repeated measures analyses on the test-scores at T2 compared to T0. Two-tailed result of t test on the GAS score and Mann-Whitney U test on the TSS score.*

	Control patients) (n=36)		**Experimental patients (n=35)**		**ANOVA**	
					Time	**Time X group**
	M, *sd*	**M T2-T0**	**M, *sd***	**M T2-T0**		
Stroop 3/2	1.6 *(0.3)*	-0.1	1.5 *(0.2)*	-0.1	n.s.	n.s.
TMT- B/A	2.2 *(1.3)*	-0.1	2.1 *(0.6)*	0.1	n.s.	n.s.
TOL nr correct	11.0 *(1.1)*	0.2	11.0 *(1.4)*	0.4	n.s.	n.s.
BADS	94.6 *(13.2)*	8.8	101.8 *(12.5)*	10.9	***	n.s.
DEX patient	25.5 *(14.6)*	-6.1	26.3 *(15.8)*	-4.3	***	n.s.
DEX proxy	28.5 *(16.6)*	-2.2	26.5 *(16.7)*	-3.6	*	n.s.
DEX therapist	28.9 *(13.5)*	-6.8	24.2 *(11.2)*	-10.6	***	n.s.
Qolibri satisfac	136.4 *(35.4)*	12.7	126.7 *(32.4)*	2.9	*	n.s.
Qolibri burden	44.1 *(12.0)*	-4.2	51.7 *(18.1)*	- 1.4	n.s.	n.s.
EOBS	22.3 *(4.2)*	1.6	25.7 *(3.5)*	4.6	***	***
RRL	7.4 *(3.2)*	-0.1	5.4 *(3.1)*	-2.2	***	***
GAS	8.9 *(3.0)*		10.7 *(2.1)*		T= 2,9	**
TSS (1-5)	3.9 *(.87)*		3.8 *(1.1)*		Z = -.4	n.s.

* p< .05 ** p< .01 ***p<.00

Table 7 shows that on T2 for most measures the effects were the same as on T1, that is, patients had neither further improved nor deteriorated. There were three exceptions: therapists' ratings on the DEX and the EOBS were significantly different for both groups. On the RRL only the experimental patients showed a significant improvement over time, indicating that they had been able to resume more role functioning activities than the control patients.

Table 8 shows the results of the *t* tests between both patient groups on the EST Totalscore, as well as on the three EST subscores (Initiative, Prospective and Executive). There were some missing data for this test, because three patients had not been able to fit this additional measure to their scheme and EST data of two other patients were unfortunately lost in the rehabilitation centre. The experimental patients performed significantly better on all scores, except for the Initiative score. In addition, results of both groups were compared to results of healthy controls. The control patients performed worse on all EST (sub)scores than the healthy controls, the experimental patients only on the EST Totalscore and the Executive score.

Table 7

Means, standard deviations and difference scores (T2-T1) of the test-variables at T2 for the control and experimental patient group. Repeated measures analyses on the test-scores at T2 compared to T1.

	Control patients (n=36)	**Experimental patients (n=35)**	**ANOVA**	
	M T2-T1	**M T2-T1**	**Time**	**Time X group**
Stroop 3/2	0.1	0	n.s.	n.s.
TMT- B/A	0.1	0.1	n.s.	n.s.
TOL nr correct	-0.3	0.1	n.s.	n.s.
BADS	1.1	0.9	n.s.	n.s.
DEX patient	-1.4	0.7	n.s.	n.s.
DEX proxy	.5	0.7	n.s.	n.s.
DEX therapist	-3.9	-1.7	*	n.s.
Qolibri satisfac	1.1	-6.9	n.s	n.s.
Qolibri burden	-1.2	2.9	n.s.	n.s.
EOBS	0.9	0.8	*	n.s.
RRL	0.2	-1.0	n.s.	*
GAS	0.3	0.2	n.s.	n.s.

* p< .05 ** p< .01 ***p<.00

Table 8

Means and standard deviations of control patients, experimental patients and healthy controls on the total score and the three subscores of the EST. Two-tailed results of t tests.

EST (range)	**CP (n=34)**	**EP (n=32)**	**HC (n=57)**	**CP vs EP** T	p	**CP vs HC** T	p	**EP vs HC** T	p
Total (0-45)	28.3(*10.2)*	34.1 *(8.2)*	37.4 *(5.6)*	2.5	*	-4.8	***	-2.3	*
Initiative (0-13)	9.5 *(3.1)*	10.7 *(2.5)*	11.5 *(1.9)*	1.6	n.s.	-3.3	**	-1.8	n.s.
Prospective (0-8)	5.2 *(2.2)*	6.8 *(1.5)*	6.7 *(1.4)*	3.4	**	-3.7	***	0.3	n.s.
Executive (0-24)	13.6 *(6.2)*	16.6 *(5.5)*	19.1 *(3.5)*	2.1	*	-4.7	***	-2.2	*

* p< .05 ** p< .01 ***p<.00

Discussion

The results of this study show that a multifaceted treatment for executive dysfunction significantly improves daily life executive functioning of ABI patients, lasting at least until six months post-treatment. The improvement was visible on a composite measure, the TEOS, in which three domains of daily life were reflected; the ability to set and accomplish realistic goals, the ability to plan, organize and regulate a series of real life tasks; and the ability to resume previous roles with respect to work, social relations, leisure activities and mobility. The effect size on the TEOS was large, and therefore clinically substantial. When considered separately, the three TEOS measures (GAS, RRL and EST) also showed significant differences between both groups. For the RRL and GAS these differences were evident after the treatment as well as at follow-up. The EST was only administered at follow-up, but also here the differences between both groups were significant for all EST scores, except for the Initiative subscore.

Our expectation that these effects would be present immediately after treatment and would subsequently remain stable at least six months after treatment was entirely met. The difference on the GAS at follow-up still indicated that the experimental patients had attained their goals to a larger extent than the control patients. Moreover, on the RRL these patients showed a higher increase than the controls at T1, while at follow-up, they had even further increased their wished-for roles in daily life, whereas the control group remained at the same activity level throughout.

In our multifaceted treatment, several elements of proven treatment methods had been incorporated, namely Problem Solving Training (Cramon von et al., 1994) and Goal Management Training (Levine et al., 2000). However, the surplus value of our treatment is its multifaceted character: a comprehensive but finite range of dysexecutive symptoms is addressed, including problems with self-awareness and self-initiative. Another distinct feature of our training is transfer to daily life situations as an integral element of the treatment. Effects of training were measured and found on indicators of EF on activity as well as participation level. In our treatment protocol the eight aspects of Ylvisaker, self-awareness, goal setting, planning, self-initiation, self-monitoring, self-inhibition, flexibility and strategic behaviour, were trained explicitly and embedded in practical exercises and home assignments. Improvements on these aspects are obvious when the measures reflecting daily life functioning are analyzed. The ability to set and accomplish realistic goals in daily life, as reflected by the GAS, depends on the capacity to be aware of one's own needs, strengths and weaknesses. In the EST, patients have to set their own goals, and make plans to organize task execution towards goals. No cues or directions are provided, so patients must initiate these tasks, carry them out and simultaneously monitor their own performance. Flexibility is necessary to adapt the execution of plans to changing circumstances and to solve possible problems, and this also requires the ability to self-inhibit actions that do not lead to the goals set. The ability to apply all these aspects on a strategic level is reflected in the RRL scores, which indicate the performance of relevant activities in daily life roles.

Before discussing the results with respect to the secondary outcome measures, we would like to stress that our study indicates that it actually is possible to treat patients with dysexecutive problems, which is not always taken for granted (Alderman, 1991). After all, dysexecutive problems are known to hamper the ability to learn new strategic behaviours and to take advantage of therapy. Nevertheless, our experimental patient group was able to adhere to the treatment; they remained motivated to follow and finish a complex, intensive, energy and time consuming treatment protocol. The results on the TSS show that these patients were satisfied with the training and the effects it had in their lives. The lower scores on the DEX patient and proxy at T1 and T2 indicate that patients, as well as significant others, experienced and observed less dysexecutive problems after treatment. However, this was also true for the control patients who experienced similar levels of satisfaction after treatment and reduction of complaints on the DEX. With respect to quality of life, both groups also showed the same pattern of results. We therefore conclude that with respect to these different indications of subjective wellbeing (satisfaction with the treatment, subjective complaints and quality of life) the effect of treatment, whether experimental or control, was the same for both groups. This result was surprising, because beforehand there were doubts about patients' motivation to perform the long and energy sapping Cogpack training. We were afraid that patients would be swiftly bored, or that they would sense that this treatment would not be effective. On the contrary, the majority of patients were very enthusiastic about the training, because Cogpack gave direct feedback on performance so that patients could monitor their improvement in the tasks over time. We therefore conclude that Cogpack training influenced patients' sense of self-efficacy positively, exactly as the experimental training did. Apparently, this has led to larger activity levels of these patients, reflected by increased role participation, although not to the same extent as the experimental patients. In addition, the control training had also positive effects on the executive abilities of trainees, as rated by therapists. On the DEX-therapist as well as on the EOBS, therapists found that both groups had significantly improved after treatment and at follow-up. However, these therapists considered the experimental patients to have improved significantly more on the DEX therapist as well as on the EOBS at both measurements.

With respect to the objective measures, there was no indication that either of the treatments had significant effects on cognitive or executive functioning as measured with neuropsychological tests. On the three conventional neuropsychological tests, Stroop, Trailmaking Test and Tower of London, there was no improvement over time at all. With respect to the memory test, included as a control measure for possible effects of the computerized training, both groups had improved on the IR score at T1. We interpret this as a re-test effect. Surprisingly, in comparison with the control group the experimental group had significantly improved on the DR score, suggesting that learning executive strategies may foster better memory performance than training memory specifically with computer exercises.

With regard to the BADS, it was also found that both groups had improved to the same extent both at T1 and T2, in comparison to baseline. This is presumably the result of a test-retest effect as well. In a previous study a considerable test-retest effect was found for the BADS (Jelicic, Henquet, & Derix, 2001). For this reason the BADS was not considered a primary outcome measure in the present study. Moreover, there is another theoretically based objection against the use of the BADS for this purpose. The aspects of novelty and problem solving are crucial elements of EF measurement. When performing an EF test repeatedly, learning effects (for instance, retaining the solution of the test problem in memory) can not be disentangled from the pure executive performance and therefore the retest assessment probably does not measure EF to the same extent again. It can therefore be seriously questioned whether the BADS, although probably more ecologically valid than other EF tests, is a sensitive measure for ED treatment effects.

Our general conclusion is that despite control patients' satisfaction and subjective wellbeing being at the same level as that of the experimental patients, the latter group did perform better on those measures that pertained to daily life executive functioning. These results prove that significant treatment effects can be accomplished by a general multifaceted treatment, if tailored to the individual patient and designed to improve activity and social participation, and that these effects last for a substantial period after ending the treatment.

Appendix I

Multifaceted treatment of Executive Dysfunction: content of the treatment protocol

The intervention protocol was based on theoretical models as well as on existing treatments. The Cognitive Schema theory of Shallice (Shallice, 1982) is the starting point. In this theory a distinction is made between schema-dictated behaviour in routine situations and controlled behaviour if schemata fall short or do not apply. In these situations a mechanism called the Supervisory Attentional System operates on selection of relevant schemata. In ABI patients with executive problems this latter mechanism is often invoked, while it is often limited in capacity or even utterly disrupted. Brouwer and Fasotti (Brouwer & Fasotti, 1997) presented an adaptation of the model of Shallice, supplemented with the important elements of self-awareness/monitoring and motivation/initiative. The multifaceted treatment is also based on the central idea of universal subgoaling, derived from cognitive architectures like SOAR (Newell, 1991) or ACT-R (Anderson, 1993). In these architectures all intelligent behaviour is seen as problemsolving behaviour and the formulation of intended actions is regarded in terms of goals and subgoals. This central notion of subgoaling was translated into a therapeutical approach, called the General Planning Approach (GPA). Furthermore, elements of Goal Management Training (Levine et al., 2000) aiming at regulation, and Problem Solving training (Cramon von et al., 1994) aiming at flexibility and problem solving, as well as more general treatment approaches of ED (as advocated by Ylvisaker) involving training of self-awareness and self-initiative, are incorporated.

The treatment is given by a neuropsychologist, if possible together with a cognitive trainer, and involves a combination of psychoeducation, strategy and skills training, and use of external devices like a diary or a PDA. In teaching the patients these skills and strategies, cognitive behavioral techniques were applied. These strategies and skills are individually tailored to a patient's specific problems, needs and goals, because the protocol can be adapted by applying variations in content and number of sessions up till a maximum of 24 session. Transfer of learning to the home situation is accomplished by using exercises and home assignments that are relevant for the subject's personal goals. The home assignments are given to the subjects at the end of every session and are extensively evaluated in subsequent sessions. The overall goal is that patients acquire an individually tailored EF strategy and have sufficient capacity for self-awareness and self-initiative to apply this strategy in daily life.

The protocol is comprised of three modules, namely 1) Information and Awareness, 2) Goal Setting and Planning, and 3) Initiation, Execution and Regulation.

Module 1, Information and Awareness, addresses Ylvisaker's aspect of self-awareness. The module has a psychoeducative character and its general purpose is improvement of awareness and the enhancement of motivation for treatment. Patients are extensively informed about dysexecutive problems and their consequences for in daily life in general, and about their own dysexecutive problems in particular. According to the model of Crosson (Crosson et al., 1989), three levels of awareness can be distinguished. Intellectual awareness can be improved by informing patients about their cognitive and executive impairments with regard to daily life functioning, in order to gain insight into their weaknesses and strengths. Furthermore, throughout the whole training, patients are continually stimulated to monitor and evaluate their own performances with the aim to improve emergent awareness. Finally, in every session patients are stimulated to predict their functioning, in order to improve anticipatory awareness, the highest level of awareness. Every next session, these predictions and their fulfilment, together with factors that did or did not help are extensively evaluated.

Module 2, Goal Setting and Planning, addresses Ylvisakers' aspects of goal-setting, planning and organizing the steps to these goals. The module is aimed at training goal setting in a systematic and structured way. Patients are taught to apply the GPA, the General Planning Approach, which allows them to formulate all (intended) activities and tasks in terms of goals and steps leading to these goals. Attention is paid to the concrete and explicit verbalization of goals in terms of when, where, with whom, with what and how long. Patients are trained to formulate concrete steps leading to a previously set goal and to put these steps in the right order. This is practised using scripts of Sirigu (Sirigu et al., 1996). Successively they learn to anticipate on eventual problems and to devise alternative steps or plans. In this module, the patient is asked to formulate three concrete goals that he/she wants to achieve by means of the treatment. These goals have to be connected with executive functioning in daily life, without other restrictions. This has resulted in a large variability in treatment goals, like for example: improving time management, being able to plan activities in advance, in order to reduce time pressure, learning to use public transport facilities in order to increase mobility, being capable of organizing activities with family or friends to improve contacts, enhancing activity in volunteer work, improving the regulation of emotions.

In module 3, Initiation, Execution and Regulation, the effective execution of plans is addressed. The module taps Ylvisakers' aspects of self-initiation, self-monitoring, self-inhibition, flexibility and problemsolving, as well as strategic behaviour. Patients are taught how to initiate execution of their plans and subsequently how to act according to plan, while constantly monitoring their performance. They learn to solve complex daily problems in a systematic way with regular checking whether higher order goals are met. In this part of the training, elements of Goal Management Training (Levine et al., 2000) as well as of Problem Solving Training (Cramon von et al., 1994) are incorporated. An important element of the training is that the patients are instructed to initiate daily life activities that involve the application of the strategies and skills that they acquire in training, since transfer is an essential element of the treatment.

Appendix 2
The Executive Secretarial Task (EST)

The EST is considered to be more ecologically valid than the usual EF tests. In this 3 hr-task a job assessment procedure is simulated. The patient is alone in a room with a box containing a series of simple secretarial assignments. On the desk there is also a list with company rules, a planning aid in the form of a day agenda, a telephone, a phonebook, a map of the floor and the location of other offices, a calculator and a small rack with office supplies. The assignments of the box have to be organized, initiated and executed, some of them with a deadline. Examples are: filling in zip codes on envelopes and posting them in time for the external post round, counting the supplies and replenishing the stocks by delivering the order form in time at the right place or searching for suitable restaurants for the company diner in the phonebook. A unique feature of this test is that, unlike most other EF tasks, it explicitly taps self-initiation. In the instruction it is only mentioned that the subject can find the assignments in the box and that they all have to be carried out. No further cues are provided on how or when the assignments have to be carried out. Indispensible materials and required information are all available, but have to be actively searched for. For example, one of the assignments is searching travel times for specific dates and destinations. This can be done by using an available telephone (and for which the instruction for use can be found in the list with company rules), but the subject is not explicitly told to use the phone for this purpose. At fixed times only, questions can be asked to a "manager". During the execution of the task, the subject is interrupted with an urgent new assignment. The task yields three scores: *Initiative*; reflecting all the actions the subject has initiated without being told so, *Prospective*; reflecting all the actions that were correctly carried out in a later stage, and *Executive*, reflecting all the actions that were correctly carried out at all. Taken together these scores form the Total score

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