The role of cognitive training in the neurorehabilitation of a patient who survived a lightning strike. A case study

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Abstract

BACKGROUND: Lightning-related injuries most often involve impairment of the functions of the central and peripheral nervous systems, usually including cognitive dysfunctions. We evaluated the cognitive deficit of a patient who had survived a lightning strike and measured the improvement after her cognitive training. This therapeutic method appears to be a powerful tool in the neurorehabilitation treatment.

OBJECTIVE: The aim of this case study was to prove the beneficial effects of cognitive training as part of the neurorehabilitation after a lightning strike.

METHODS: Six neuropsychological functions were examined in order to test the cognitive status of the patient before and after the 2-month cognitive training: phonological short-term memory (digit span test and word repetitions test), visuo-spatial short-term memory (Corsi Block Tapping Test), working memory (backward digit span test and listening span test), executive functions (letter and semantic fluencies), language functions (non-word repetition test, Pléh-Palotás-Lőrik (PPL) test and sentence repetition test) and episodic memory (Rivermead Behavioral Memory Test and Mini Mental State Examination). We also utilized these tests in aged-matched healthy individuals so as to be able to characterize the domains of the observed improvements more precisely.

RESULTS: The patient exhibited a considerable improvement in the backward digit span, semantic fluency, non-word repetition, PPL, sentence repetition and Rivermead Behavioral Memory tests.

CONCLUSIONS: The cognitive training played an important role in the neurorehabilitation treatment of this lightning injury patient. It considerably improved her quality of life through the functional recovery.

Keywords: lightning injury, neurorehabilitation, neuropsychological tests, cognitive training

1. Background

Lightning is one of the major weather-related phenomena that cause serious health injuries and death. Although the principal cause of death in these cases is cardiac arrest, most of the complications faced by the survivors are due to the injuries to the nervous system, which can result in temporary or permanent disabilities (reviewed in Cherington, 2003, 2005; Janus & Barrash, 1996). Lightning-related neurologic syndromes can be classified into four categories. The first includes immediate and transient symptoms, which generally disappear within a few days. These symptoms mainly comprise a loss of consciousness, amnesia, confusion, headache, paresthesia and weakness (e.g. keraunoparalysis). The MRI of the skull often reveals cerebral edema (Cherington, Yarnell, & Hallmark, 1993), while the EEG findings commonly include diffuse abnormal activity characterized by a slowing or poorly organized background rhythm with low amplitude (E. Muller, Endler, & Wedekind, 1978). The second group includes immediate and prolonged or permanent symptoms due to post-hypoxic-ischemic encephalopathy (a complication of cardiac or respiratory arrest), cerebral infarction, intracranial hemorrhages, a cerebellar dysfunction, myelopathy, dysfunctions of the autonomic nervous system, peripheral nerve lesions and myopathy. This category also involves neuropsychological impairments, which can be subcategorized as physical (headache, chronic fatigue, light and sound sensitivity, sleep disturbance and musculoskeletal pain), emotional (irritability, lability, anxiety, depression with a feeling of helplessness, anhedonia and loss of self-confidence) or cognitive (attention, memory, problem-solving, executive functions and coordination, and psychomotor speed) dysfunctions. (reviewed in Primeau, 2005; van Zomeren, ten Duis, Minderhoud, & Sipma, 1998; Yarnell, 2005; Frayne & Gilligan, 1988). The third group comprises delayed neurologic syndromes, such as amyotrophic lateral sclerosis (Jafari, Couratier, & Camu, 2001), movement disorders (O'Brien, 1995) and psychiatric symptoms such as post-traumatic stress disorder (Cherington, Yarnell, & Lammereste, 1992), which occur days to months following the lightning strike. The fourth group includes the lightning-linked secondary trauma resulting from falls. In consequence of the lightning strike-related dysfunctions that emerge in the latter three categories, neurorehabilitation, including cognitive rehabilitation techniques, is crucial in most of the patients.

2. Objective

The aim of this case study was to prove the beneficial effects of neurorehabilitation after a lightning strike, focusing on the changes in the cognitive functions after the early, personalized cognitive rehabilitative training.

3. Methods

3.1. Case history and patient examination

The subject was a 40-year-old female who had been struck by lightning during outdoor activity. After a cardiac arrest, prompt cardiopulmonary resuscitation was implemented. Hyperemia was observed on the left shoulder and the left side of the neck. The EEG recorded on the second day after her admission to the intensive care unit of the Health Center of the Hungarian Defense Forces, revealed diffuse slow wave activity. The improving respiratory functions and recovery of consciousness allowed a head MRI on the fifth day of hospitalization. This showed slight multifocal T2 hyperintense signals subcortically in the left frontal lobe and bilaterally in the parieto-occipital regions, and also cerebellar edema. On the eleventh day, the patient was transferred to our Department of Neurology. On neurological examination, the muscle strength was proximally 2/5 and distally 4/5 in the left arm, and 4/5 in the other limbs. Hyporeflexia (+) was noted as regards the deep tendon reflexes. The bilateral Babinski sign can be observed. The neck muscles were tender without signs of meningeal irritation. The patient was unable to stand or walk because of the severe cerebellar ataxia. Moderate motor aphasia was observed. The patient was disoriented in time and space, and displayed retrograde and anterograde amnesia, slowed psychomotor activity and sometimes agitation. The thinking was occasionally disorganized and incoherent, with disruptions and blocking. Delusional fragments and severe attention deficit were also noted. The facial expressions and body movements reflected moderate pain. The sleep was disrupted by several awakenings. Elevated serum lactate dehydrogenase and creatine phosphokinase levels were measured. On the eighth day after admission, the skull MRI did not indicate cerebellar edema, but perivascular white matter lesions were still observed in the left hemisphere. The EEG demonstrated transient slowing in the left temporal lobe. On the following day, the electroneurography of the median and ulnar nerves, and also the medial antebrachial cutaneous nerves, showed no abnormalities. Lesions of the peripheral nerves could therefore be ruled out as a cause of the left limb weakness. Immediately after the period of hospitalization in the acute ward, the mobilization of the patient was started by a physiotherapist. The patient was transferred to our neurorehabilitation unit on the 23rd day after the lightning strike. At that time her cooperation and orientation were good, the periods of amnesia were shorter and there were no signs of agitation. Deep tendon reflexes could not be elicited in the upper limbs, whereas they were exaggerated in the lower limbs (+++). The Babinski sign could not be evoked on either side. A slight proximal weakness remained in the left arm, but otherwise the muscle strength was regained. Slight ataxia was noted in the right arm and severe ataxia in the legs, with gait ataxia of sensory type. The speech was not affected.

3.2. Methods of neurorehabilitation

In the neurorehabilitation unit, the patient received cognitive training, physiotherapy (therapeutic exercise) and occupational therapy for 2 months.

3.2.1. Assessment of cognitive functions

3.2.1.1. Subjects

The patient and 6 healthy control participants were matched for age, education and gender. Three of the normal control subjects took part in the cognitive therapy and their cognitive functions before and after the therapy were compared with those of the patient and those of the 3 control individuals who did not take part in cognitive training. All participants were fully informed in advance about the methods and aims of the study and provided their written informed consent to participation.

3.2.1.2. Neuropsychological tests

For cognitive function testing, 6 major neuropsychological functions were examined in both the patient and the control subjects before and after the 2 months with or without cognitive training: phonological short-term memory, visuo-spatial short-term memory, working memory, executive functions (verbal fluency - letter and semantic), language functions and episodic memory.

3.2.1.2.1. Phonological short-term memory

The phonological short-term memory was measured with a digit span test and a word repetition test. The digit span test involves the spoken presentation of sequences of digits for immediate serial recall. Four sequences are presented for each length, starting with 3-digit sequences. If 3 of the 4 sequences of a particular length are correctly recalled, the sequence length is increased by one. One digit is presented per second, and the maximum length at which 3 sequences are correctly recalled provides a measure of the digit span. The word repetition test is similar to the digit span test, but in this task the subject has to repeat 2 syllables or morphologically complex words in the same sequence as presented to him/her. No words are repeated. The length of the sequence is increased by one after at least 3 successful repetitions out of 4 attempts (see Racsmány, Lukács, Németh, & Pléh, 2005).

3.2.1.2.2. Visuo-spatial short-term memory

The visuo-spatial short-term memory was measured by the Corsi Block Tapping Test (Lezak, 1995). In this nonverbal task, 9 cubes were presented on a black board and the subject has to touch the cubes in the same sequence as the experimenter. The length of the sequence is increased by one after at least 3 successful repetitions out of 4 attempts.

3.2.1.2.3. Working memory

The working memory was measured with the backward digit span and listening span tests. The backward digit span test involves the same procedure as the digit span and word repetition tests, but in this task the participant has to recall the digits in the reverse sequence. The working memory capacity was further measured with the Hungarian version of the listening span task (Daneman & Blennerhassett, 1984). In this test, the experimenter reads aloud increasingly longer sequences of sentences and the subject has to make a “true” or “false” decision concerning the sentence and then recall the final word in each of the sentences in sequence. The working memory capacity of the subject is defined as the longest sequence length at which the final words are successfully recalled (see Janacsek, Tánczos, Mészáros, & Németh, 2009; Racsmány et al., 2005).

3.2.1.2.4. Executive functions - Verbal (letter and semantic) fluency tasks

In letter fluency tests, in a limited period of time (60 seconds), the participants have to recall as many words as they can that begin with a certain letter, e.g. *F, A* or *S* (in English) or *K, T* or *A* (in Hungarian). They are asked to avoid repetitions and proper names (Benton, Hamsher, & Sivan, 1976; Raskin, Sliwinski, & Borod, 1992; Troyer, Moscovitch, & Winocur, 1997; Troyer, Moscovitch, Winocur, Alexander, & Stuss, 1998; Troyer, Moscovitch, Winocur, Leach, & Freedman, 1998; Troyer, 2000). In semantic fluency tests, the words must belong in a specified semantic category, e.g. fruits or animals, and the participants must recall as many words as they can in a limited period of time (Cardebat, Doyon, Puel, Goulet, & Joanette, 1990; Mattis, 1988).

3.2.1.2.5. Language functions

In non-word repetition test, the participants must repeat longer and longer meaningless words after hearing them. Non-words correspond to the phonological construction of the participant's native language. Because of the need for an immediate repeat, there is no chance for reiteration and articulatory bracing; this task therefore involves phonological storage (Németh, Racsmány, Kónya, & Pléh, 2001). PPL: Pléh, Palotás and Lőrik constructed this test in order to measure the state of development of language inflexions, coding and spatial relations, to analyze the use of postpositions and to deduce the level of maturity of the language (Németh et al., 2012; Pléh, Palotás, & Lőrik, 2002). In the sentence repetition task, the participants repeat meaningful sentences after hearing them. This task is often used to assess aphasic and other patients in neuropsychological settings.

3.2.1.2.6. Episodic memory

We applied the Rivermead Behavioral Memory Test, which contains both verbal and visual recognition tasks and delayed evocation tasks, and the Mini Mental State Examination

(MMSE), in which orientation, attention, executive functions and the ability to make rapid associations are assessed.

3.3. Rehabilitation program

The patient participated in a personalized therapeutic protocol in the cognitive rehabilitation program in order to improve her cognitive deficits. The 3 subjects of one of the control groups took part in the same 1x60 minute cognitive training program every day. This protocol involved the improvement of logical and attentional skills (with stimulus and word seeking tasks, which involved searching for words and figures hidden among numerous stimuli/letters), executive functions and short-term memory (word list learning, digit and backward digit span). We strived to improve the poor verbal short-term memory of the patient with visualization techniques too. The first technique was to remember name-face relations: the patient had to make a visual association between the name and specific characteristics of the face. The other visualization technique was the use of an agenda: the patient had to consult the agenda at several fixed moments during the day (Ponds & Hendriks, 2006). For improvement of the visuo-spatial memory, we used tasks in which the subject visualized figures featuring on a map from memory. To improve the complex verbal working memory, we used training tasks where the information-holding and processing areas were activated at the same time: repeated performance of verbal working memory tasks, with feedback and rewards based on the accuracy for every trial and explicit rehearsal techniques or meta-cognitive strategies (Klingberg, 2010). We also used tests of reading comprehension, recognition of spelling errors and phonological reading ability (Dahlin, 2011). We improved the executive functions with learning of self-regulatory and/or meta-cognitive strategies that are helpful in a variety of situations, such as interrupting an ongoing task and reflecting on one’s intentions (Boelen, Spikman, & Fasotti, 2011). We additionally used memory supporting tasks for everyday life, e.g. making to-do lists, keeping a diary, or learning how to use the reminder function on a cell phone. After every session, the participants received homework to be presented in the next session.

The patient was also given therapeutic exercise for 45 minutes per day in order to enhance her muscle strength and to improve her physical condition, coordination and walking. She participated in daily occupational therapy lasting for 45 minutes so as to regain fine motor skills and activities of daily living.

3.4. Data analysis

The SPSS 15.0 software was utilized for descriptive statistical analyses of the achievements of the two control groups (treated and not treated groups) in order to compare their mean performances both with each other and with that of the patient. As we were assessing only a single patient, analysis of variance could not be used for the group and patient comparisons. The extent of the difference was indicated by using the mean test values of the control groups with and without cognitive rehabilitation.

4. Results

4.1. Phonological short-term memory

Digit span task. The performance of the patient in the digit span short-term memory task was similar to that of the control groups, and the performance was not affected at all by the cognitive training, i.e. the patient's capabilities were the same as those of the controls at the beginning and at the end of the treatment or observation period (not trained) (Table 1).

Word repetition test. The performance of the patient was again similar to the results of those control groups. The cognitive training did not lead to an improvement (Table 1).

4.2. Visuo-spatial short-term memory

Corsi Block Tapping Test. The performance of the patient was approximately 1 unit lower than those of the control groups. The difference remained the same after the cognitive training (Table 1).

4.3. Working memory

Backward digit span test. The performance of the patient was more than 1 unit poorer than those of the control groups with or without cognitive training (Table 1). Improvements were noted for the patient and the control group with cognitive training, but not for the group without any training (Figure 1).

Listening span tasks: The performance of the patient was rather similar to that of the control groups; the cognitive training did not affect the test results (Table 1).

4.4. Executive functions - Verbal (letter and semantic) fluency tasks

The performance of the patient in the verbal fluency tasks was considerably poorer (a difference of more than 4 words in the letter fluency task and of more than 2 words in the semantic fluency task) than those of the control groups (Table 1). Although the cognitive training resulted in a slight improvement in the letter fluency task in the control subjects, the patient did not exhibit any improvement of this function. However, in the semantic fluency task, the performance of the patient improved considerably (Figure 1).

4.5. Language functions

In all the applied tests of language functions, the performance of the patient was again appreciably poorer than those of the control groups (a difference of approximately 4 units in the non-word repetition task, and of 2 mistakes in the PPL and sentence repetition tasks; Table 1). Thanks to the cognitive training, the performance of the patient improved in all of these tasks, and the control subjects too demonstrated a slight improvement in the non-word repetition task (Figure 1).

4.6. Episodic memory

In the Rivermead Behavioral Memory Test, the performance of the patient was 3 units lower than those of the control groups, while there was no major deficit in the MMSE (Table 1). The cognitive training practically eliminated the previously observed dysfunction in episodic memory (Figure 1).

4.3. The result of physical and occupational therapy

After the 2-month rehabilitation period, the patient's neurological symptoms had almost disappeared. The muscle strength was generally 5/5. There was still a slightly ataxic gait. The patient was able to walk independently, and she regained her self-sufficiency.

5. Discussion

A lightning strike can result in both acute and chronic serious health impairments. The injuries involving the greatest level of deterioration are those affecting the functioning of the central and peripheral nervous systems. As regards the central nervous system injuries, the impairment of the cognitive functions is sometimes a somewhat neglected aspect of the neurological dysfunctions. However, such cognitive function impairments can worsen the quality of life to a great extent. Thus, before the rehabilitation training the neuropsychologist has the important role of measuring the range of the neuropsychological deficits. Thereafter, through the application of various cognitive rehabilitation techniques, improvements can be attained in the cognition and the functions of information processing (via memory, executive functions, language functions and problem solving strategies). The main goal of this cognitive training is to achieve an improvement in the patient's functionality in everyday life. The present case is an example illustrating that this goal can be attained.

The assessment of cognitive functions makes use of numerous tests in which an attempt is made to localize the brain areas responsible for the impaired mental functioning. Phonological short-term memory includes two main components: an input storage system (the phonological short-term store) and an output rehearsal process. These components are bound to discrete parts of the left cerebral hemisphere (Vallar, 2006; Paulesu, Frith, & Frackowiak, 1993; Smith, Jonides, Marshuetz, & Koeppe, 1998). Visuo-spatial short-term memory tasks depend on intact dorsal parietofrontal networks (Croize et al., 2004), with activation in the right occipital gyrus (Dupont et al., 1993), the right posterior parietal gyrus (Belger et al., 1998), the right premotor area and the right inferior dorsolateral prefrontal cortex. The prefrontal cortex has a crucial role in the working memory (N. G. Muller & Knight, 2006). Most working memory tasks recruit the network of the prefrontal cortex and parietal areas (Honey et al., 2002). For intact executive functions, both frontal (particularly the prefrontal cortex) and non-frontal (temporal) brain regions seem to be necessary (Fuster, 2000). As regards language functions, the non-word repetition task mainly loads the store component of the phonological loop and not the rehearsal component (Baddeley, Gathercole, & Papagno, 1998). In the sentence repetition test, predominantly the verbal working memory is used. The cerebellum appears to be involved in the modulation of a wide spectrum of linguistic functions, such as verbal fluency, word retrieval, syntax, writing, reading and metalinguistic abilities (Murdoch, 2010). The right anterior and posterior prefrontal regions play essential parts in episodic memory retrieval (Nyberg, 1998). The medial temporal lobe, including the hippocampal formation itself, also has a fundamental role in episodic memory (Eldridge, Knowlton, Furmanski, Bookheimer, & Engel, 2000; Schacter & Wagner, 1999; Vargha-Khadem et al., 1997).

The patient initially displayed impairments of several of the cognitive functions listed above: her performance at that time was unsatisfactory in the backward digit span, verbal fluency tasks, language function tests and Rivermead Behavioral Memory Test as compared with the performance of the control subjects. In brief, she had slight problems with her working memory (prefrontal cortex and parietal areas), and considerable problems with her executive functions (frontal and temporal lobes), language functions (fronto-temporal, parietal and cerebellar regions) and episodic memory (right anterior and posterior prefrontal regions, and the medial temporal lobe, including the hippocampal formation). The presence of these cognitive abnormalities generally correlated with the structural and functional abnormalities reflected in the MRI scans (a hyperintense signal frontally and subcortically in the left hemisphere, and bilateral parieto-occipital and cerebellar edema at the first imaging, and slight vascular white matter lesions in the left hemisphere on the second occasion). The EEG recordings (diffuse slowing initially and transient slowing in the left temporal lobe on the second occasion) also reflected the dysfunctioning of these injured areas.

Cognitive rehabilitation involves two basic strategies (Park & Ingles, 2001). One comprises the retraining of the cognitive processes impaired by the damage to the neural circuits. These cognitive processes can be retrained if they were not irreparably lost as a result of the injury (Robertson & Murre, 1999). The second strategy is to attempt to find new compensatory skills to improve the performance in everyday tasks. The patients have to discover how they can compensate their deficits with newly-learned strategies, through the use of preserved cognitive skills and functional reorganization of the brain (Backman & Dixon, 1992; Vanderploeg et al., 2006). As concerns the effects of cognitive rehabilitation, it has been found (Feinstein, Brown, & Ron, 1994) that healthy subjects have the ability to improve their performance significantly with practice in tests of attention. It has been reported that memory (including working memory) could be improved by training (Verhaeghen, Marcoen, & Goossens, 1992; Olesen, Westerberg, & Klingberg, 2004).

After the cognitive training, the performance of our patient was improved in the tests of backward digit span (working memory), semantic fluency (executive functions), non-word repetition, PPL and sentence repetition (language functions) and the Rivermead Behavioral Memory Test (episodic memory). There are a number of factors that can influence the achievement of this kind of improvement, e.g. motivation, the effects of practice on the results of the tests, the spontaneous recovery of cognitive functions during the rehabilitation period and training-induced plasticity (Rohling, Faust, Beverly, & Demakis, 2009). These tests should therefore also be performed on an accompanying group of appropriately selected healthy subjects in order to characterize the domains of the observed improvements more precisely. Accordingly, we recruited 6 age-matched healthy controls, 3 of whom received cognitive training, and 3 of whom did not. With this study setup, we assessed the spontaneous improvement during the 2 months between the first and second examinations (control subjects without cognitive therapy) and the therapy-induced improvement in cognitive functions under normal conditions (control subjects with cognitive therapy). The control examinations did not reveal any significant spontaneous improvements in the tests. It is important to mention that the control subjects had already reached the maximal score in the tests of PPL, sentence repetition and MMSE. However, the cognitive training did lead to considerable improvements in the backward digit span and letter fluency tasks in control subjects. The patient demonstrated the most marked improvements in the tests of semantic fluency (temporal lobe), non-word repetition, PPL and sentence repetition (fronto-temporal, parietal, cerebellar regions) and the Rivermead Behavioral Memory Test (right anterior and posterior prefrontal regions, the medial temporal lobe including the hippocampal formation). The explanation of these findings is that the areas that suffer more severe (but not irreversible) injury and dysfunction can improve more extensively than the slightly injured ones in consequence of the cognitive training-induced synaptic plasticity (Lovden, Backman, Lindenberger, Schaefer, & Schmiedek, 2010).

Both the physical and the occupational therapy applied for neurorehabilitation had beneficial effects: the patient's motor symptoms almost disappeared and her daily activities and skills virtually normalized. She became self-sufficient, and her quality of life returned to almost the same level as before the lightning strike.

This single case study provides the first detailed report of the beneficial effects of cognitive therapy as an important part of the neurorehabilitation after an injury caused by a lightning strike.

6. Declaration of interest

The authors declare no conflicts of interest.

7. Acknowledgments

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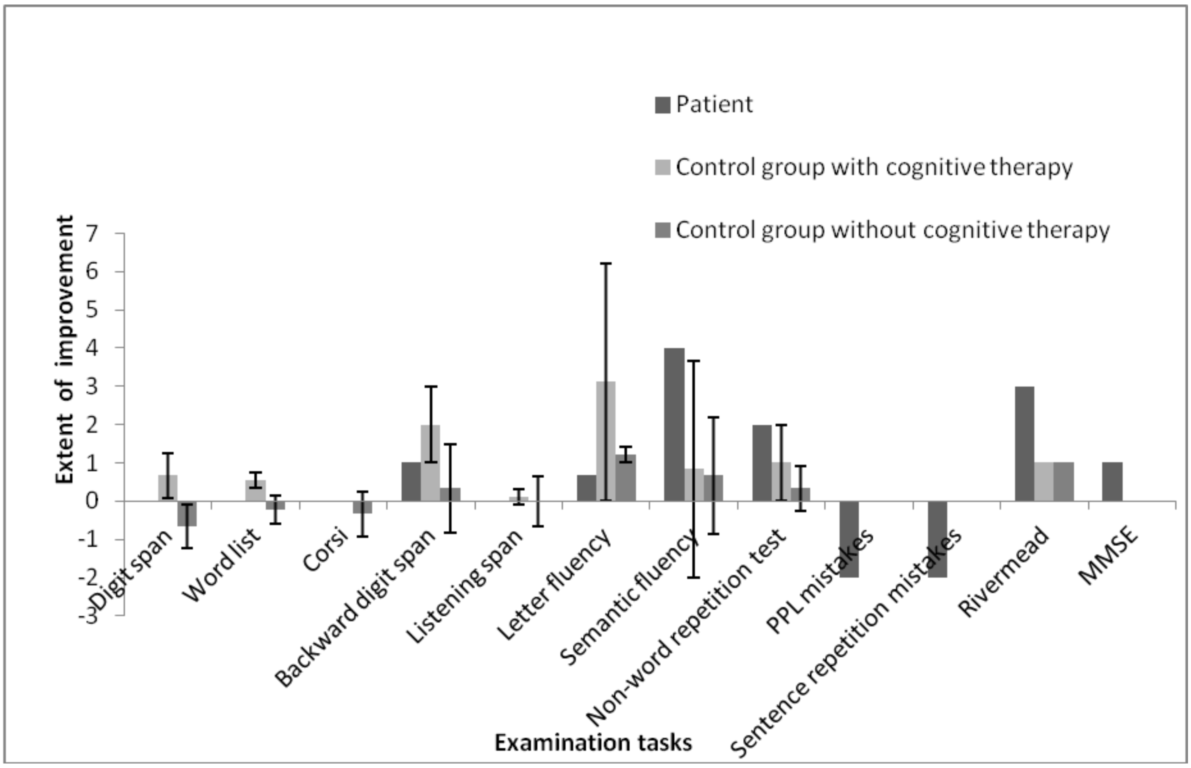
Table 1. The performances of the patient and the control subjects in the different tests of cognitive functions on the two occasions

	Initial tests			Second tests			Differences between second and initial tests		
	Patient	Control group with training	Control group w/o training	Patient	Control group with training	Control group w/o training	Patient	Control group with training	Control group w/o training
Digit span	5	5.33 (0.57)	5.66 (0,57)	5	6 (1)	5 (0)	0	0.66 (0.57)	-0.66 (0.57)
Word repetition	4.33	3.77 (0.38)	3.55 (0.5)	4.33	4.33 (0.33)	3.33 (0.57)	0	0.55 (0.19)	-0.22 (0.38)
Corsi Block Tapping	4	5 (0)	4.66 (0.57)	4	5 (0)	4.33 (0.57)	0	0 (0)	-0.33 (0.57)
Backward digit span	2	3.66 (0.57)	3.66 (0.57)	3	5.66 (0.57)	4 (1)	1	2 (1)	0.33 (1.15)
Listening span	3	3.66 (0.57)	3.11 (0.84)	3	3.77 (0.38)	3.10 (0.69)	0	0.11 (0.19)	-0.003 (0.66)
Letter fluency	7.66	13.33 (3.52)	11.77 (0.76)	8.33	16.44 (3.09)	12.99 (0.57)	0.67	3.11 (3.09)	1.22 (0.19)
Semantic fluency	16	22.16 (2.56)	18.16 (2.36)	20	23 (2.64)	18.83 (2.84)	4	0.83 (2.64)	0.66 (1.52)
Non-word repetition	3	6.66 (0,57)	7 (1)	5	7.66 (0.57)	7.33 (0.57)	2	1 (1)	0.33 (0.57)
PPL mistakes	2	0 (0)	0 (0)	0	0 (0)	0 (0)	-2	0 (0)	0 (0)
Sentence repetition mistakes	2	0 (0)	0 (0)	0	0 (0)	0 (0)	-2	0 (0)	0 (0)
Rivermead Behavioral Memory	5	8 (0)	8 (0)	8	9 (0)	9 (0)	3	1 (0)	1 (0)
MMSE	29	30 (0)	30 (0)	30	30 (0)	30 (0)	1	0 (0)	0 (0)

8. Figure captions

Figure 1. The extent of improvement after 2 months with or without cognitive training


Patient
Control group with cognitive therapy
Control group without cognitive therapy
Extent of improvement
7
6
5
4
3
2
1
0
-1
-2
-3
Digit span
Word list
Corsi
Backward digit span
Listening span
Letter fluency
Semantic fluency
Non-word repetition test
PPL mistakes
Sentence repetition mistakes
Rivermead
MMSE
Examination tasks