Verbal short-term store-rehearsal system and the cerebellum Evidence from a patient with a right cerebellar lesion

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Summary

We describe an 18-year-old patient who underwent surgical removal of the right cerebellar hemisphere for the presence of a neoplastic lesion. After surgery, the patient's neuropsychological examination was normal except for a transient selective verbal short-term memory (STM) impairment characterized by reduced verbal digit span and rapid forgetting of verbal material. An extensive examination of the patient's deficit was performed in order to identify which of the two components of STM (phonological short-term store and/or rehearsal system) was impaired. The functional locus of the deficit was identified at the level of the phonological output buffer, a component of the rehearsal system, as suggested by the pattern of results obtained, namely: the improvement of Correspondence to: Dr Maria Caterina Silveri, Neuropsychology Service, Institute of Neurology, Catholic University, Largo A. Gemelli 8, 00168 Rome, Italy. E-mail: iclnp@unicatt.rm.it

the digit span seen with pointing compared with the verbal response; the advantage of auditory over visual presentation of digits; and the lack of a phonologicalsimilarity effect with visual presentation of letters. On the other hand, the functioning of the phonological store was demonstrated by the normal amplitude of the recency effect in free recall of words and by the phonologicalsimilarity effect with auditory presentation of letters. Our finding is consistent with previous functional (PET) studies showing the involvement of the right cerebellum during tasks requiring silent recirculation of verbal information. We conclude that the cerebellum takes part in the planning of speech production at a level that does not require an overt articulation.

Keywords: verbal short-term memory; rehearsal process; cerebellum

Abbreviations: STM = short-term memory; ph-STS = phonological short-term store

Introduction

In the last 15 years many experimental studies, both on patients with cerebral lesions and on normal subjects, have been devoted to understanding the functional organization of the verbal (or phonological) short-term memory (STM) system (Baddeley, 1986, 1990), i.e. of the memory function that underlies the retention of verbal information over a short period of time. This memory system, also called the phonological loop, is supposed to take part in specific cognitive abilities such as sentence comprehension (Warrington et al., 1971; Saffran and Marin, 1975; Clark and Clark, 1977; Shallice, 1979; Baddeley et al., 1987; Baddeley and Wilson, 1988), new word (therefore new language) acquisition (Baddeley et al., 1988; Papagno and Vallar, 1992, 1995) and word repetition (Martin, 1996). The phonological loop does not have unitary functional architecture but comprises two independent subsystems or components

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(Salamè and Baddeley, 1982). The phonological short-term store (ph-STS) is a limited capacity store in which the verbal information is passively held in phonological form only for a limited period of time, and the rehearsal system is a subarticulatory process which recirculates stored phonological information in order to prevent its rapid decay. If verbal material is presented in spoken form its access to the ph-STS is direct and obligatory. If verbal material is presented visually (written stimuli or pictures) then it needs to be recoded into a phonological code by means of the rehearsal process before the ph-STS is accessed (Sperling, 1963; Levy, 1971). Thus, the rehearsal process has two functions: refreshing phonological traces which otherwise fade and translating visual stimuli into phonological code. The recirculation of the memory trace provided by the rehearsal system is supposed to take place between the ph-STS and

the so-called phonological output buffer (for a discussion, see Shallice and Vallar, 1990), a working memory space in which the phonological segments are temporarily stored, in order to allow the programming of various output processes (Burani *et al.*, 1991).

Converging evidence from pathology (Vallar and Papagno, 1995; Vallar *et al.*, 1997) and PET activation studies in normal subjects (Paulesu *et al.*, 1993) suggests that STM components have discrete neural correlates; the left supramarginal gyrus and the left inferior parietal lobe are crucial areas for the functioning of the ph-STS, whereas the left inferior frontal gyrus (Brodmann's area 44, i.e. Broca's area), the left premotor cortex, the insula and the supplementary motor area are crucial for the rehearsal system. Interestingly, PET studies in normal subjects also show that experimental tasks involving the rehearsal system, such as rhyming judgements for visually presented letters, produce asymmetric activation of the cerebellum, with prevalent activation of the right cerebellum (Paulesu *et al.*, 1993).

We describe a patient with a neoplastic lesion of the right cerebellum who presented a selective impairment of verbal short-term memory. Recent papers have demonstrated the involvement of the cerebellum in cognitive processes traditionally attributed to supratentorial structures (Silveri *et al.*, 1994; Schmahmann, 1996; Schmahmann and Sherman, 1998; Molinari *et al.*, 1997; Silveri *et al.*, 1997). In the present report we offer further evidence in support of this view and demonstrate that the cerebellum has a role in verbal STM functioning. An extensive investigation of our patient's verbal STM has allowed definition of the precise locus of the deficit that we attribute to the impairment of the rehearsal component, in agreement with results obtained in functional studies.

Case report

An 18-year-old, right-handed boy with 11 years of formal education presented a 2-week history of intense occipitalfrontal headache which was unresponsive to pharmacological treatment, complicated by vomiting and imbalance. He was admitted to the Neurosurgery Department of the Catholic University in January, 1997. CT and MRI showed an isodense, 48×40 mm mass in the right cerebellar hemisphere with minimal involvement of the posterior vermis, causing a mild mass effect on the fourth ventricle and dilatation of the supratentorial ventricles. The tumour presented an irregular morphology with areas of necrosis and intense enhancement after administration of intravenous contrast medium. The patient was co-operative and orientated in time and place. Physical examination revealed wide-base, ataxia of gait and stance, right lateropulsion and right dysmetria in finger-tonose and heel-to-knee tests. Funduscopy showed bilateral papilloedema. The rest of the neurological examination was normal. On February 2, 1997 the patient underwent surgical removal of the mass. The tumour was approached using suboccipital craniotomy on both sides with right prevalence.

A complete resection of the lesion, which proved to be a medulloblastoma with desmoplastic focal features, was achieved. The post-operative period was normal. Physical examination still showed wide-base, ataxia of gait and stance, and right dysmetria in finger-to-nose and heel-to-knee tests. Slight hypotonia of the right limbs, palsy of the right seventh cranial nerve, right hypoacusia and slight hypoaesthesia in the territory of the right trigeminal nerve were also present. Eye movements were normal except for nystagmus in lateral and up-down gaze and hypometria of saccades. Cerebellar motor impairment was quantified by using a modified version of the motor deficit scale proposed by Appollonio et al. (1993), which ranges from zero (absence of any deficit) to 42 (presence of all deficits to the highest degree). Our patient reached a total impairment score of 11. No sign of dysarthria was found (score = 0) (dysarthria scale ranges from zero to 4; see Appollonio et al., 1993). A post-operative MRI confirmed the surgical excision of the posterior right cerebellar lobe and, to a lesser extent, of the posterior vermis; the right anterior cerebellar lobe and the nucleus dentatus were spared (Fig. 1). There was no evidence of supratentorial or spinal lesions. A clinical check-up was performed in July, 1997. The physical examination was completely normal. The patient gave informed consent to the tests. He received a detailed written report of the results for use in clinical rehabilitation.

General neuropsychological examination

A general neuropsychological examination was performed shortly before surgery. The patient was oriented in time and in place and was co-operative. It is worth noting that the immediate and delayed recall of words was completely normal (raw scores 51/75 and 11/15, adjusted scores 42.9 and 8.2 and cut-off scores 28.53 and 4.69, respectively). No sign of memory disorders was detected except for a reduced verbal and spatial span (verbal span = 4; spatial span = 4). Oral, limb and constructional praxis were normal, as was visuospatial analysis. Intelligence and frontal lobe functions were spared. Spontaneous speech was normal; no linguistic deficit was detected in production tasks except for a moderately reduced verbal fluency. Comprehension was good. Reading and repetition were normal. Handwriting was consistent with moderate peripheral dysgraphia, but spelling was good. The patient was checked again a few days after the surgery. Spatial span and verbal fluency had returned to normal, although a very mild peripheral dysgraphia persisted. The verbal span was unmodified. An extensive 3-day evaluation of the patient's short-term memory was performed at that time (starting 3 days after surgery). A neuropsychological check-up was made in July, 1997. The patient's scores were normal on all tasks, including those of the short-term memory battery. The patient's scores, adjusted for age and education, on the three general neuropsychological examinations are summarized in Table 1. From the experimental testing we reported the patient's results on the

various experimental short-term memory tasks 3 days after the surgery (February 1997) and 5 months later (July 1997).

Methods

General assessment of STM functioning

Immediate span

The traditional way to assess STM functioning is the immediate span, which requires the immediate serial recall of lists of verbal stimuli (digits, letters, words) in their presentation order. The score is the length of the longest correctly recalled sequence. Serial recall of verbal stimuli is generally explored in the auditory verbal modality (repetition) but stimuli may also be administered in the visual modality (by presenting series of numerical digits, each written on a card) and recalled by pointing to digits displayed on a card. Normal subjects usually present greater span with verbal stimuli than with visual stimuli (Conrad, 1964; Levy, 1971) suggesting that the store primarily responsible for span is especially useful for auditory presentation of verbal material (Shallice and Vallar, 1990). Conversely, better performance with visually presented stimuli is a typical feature of patients with STM deficit (but some exceptions do exist; for a discussion, see Shallice and Vallar, 1990), making the existence of a visual short-term store separate from the auditory short-term store plausible (Margrain, 1967; Sperling, 1967). Patients with impaired STM generally do not show any advantage with a pointing procedure when compared with a verbal modality of recall (Warrington and Shallice, 1969). A definite improvement in immediate memory scores by use of the pointing procedure has only been described in a few patients (Kinsbourne, 1972; Romani, 1992). The immediate memory advantage of the pointing procedure has been taken as evidence of a reduced capacity of the phonological output buffer, a component of the rehearsal system (Vallar et al., 1997).

Forgetting

Another general feature of the STM deficit is rapid forgetting of verbal material (within a few seconds) when the patient is engaged in the verbal interference activity of counting backwards compared with normal subjects in whom, conversely, forgetting is negligible (Brown, 1958; Peterson and Peterson, 1959).

As already mentioned, the STM system comprises two components: the ph-STS and the rehearsal process. Each component may be selectively explored by means of appropriate procedures, namely, the phonological-similarity effect and the word-length effect.

Phonological-similarity effect

Immediate serial recall of lists of phonologically dissimilar stimuli in auditory presentation is greater than recall of phonologically similar stimuli (stimuli may be letters or words) (Conrad, 1964; Wickelgren, 1965; Baddeley, 1966). The presence of phonological interference suggests that information is held in the ph-STS according to a phonological code. Thus, the absence of the phonological-similarity effect could be traced back to a deficit at the level of the ph-STS. In normal subjects, the phonological-similarity effect is also observed when stimuli are presented visually. This is taken as evidence of normal recoding of visual information into a phonological code by means of articulatory rehearsal. The presence of the phonological-similarity effect for auditory presentation, but not for visual presentation is thus consistent with a normal ph-STS and a defective rehearsal process.

Word-length effect

The immediate serial recall of lists of words is greater for short than for long words, with either auditory or visual presentation (Baddeley *et al.*, 1975). The word-length effect may be interpreted by metaphorically describing the rehearsal system as a time-limited loop which recirculates more short words than long ones (Baddeley *et al.*, 1975). For this reason, the word-length effect can be considered a good indicator of the efficiency of articulatory rehearsal.

Ph-STS versus rehearsal deficit

Appropriate tasks generally allow discrimination of a deficit at the level of the ph-STS from an impairment of the rehearsal process. As already mentioned, patients with selective impairment of the ph-STS present superior span performance with visual presentation of stimuli compared with auditory presentation (in normal subjects the opposite pattern is observed). The possibility that the superiority of visual over auditory span in patients with impaired STM might be due to an impaired phonological decoding of the verbal stimulus is ruled out by the observation that in these patients the ability to repeat single stimuli correctly is typically preserved.

In a smaller group of patients, the deficit has been placed at the rehearsal level. A typical feature in these cases was the absence of the phonological-similarity effect for visually presented stimuli and of the word-length effect. Indirect evidence of the functioning of the rehearsal process is the effect of articulatory suppression; the continuous uttering of irrelevant speech is supposed to interfere with the activity of the rehearsal processing of verbal stimuli (with both auditory and visual presentation), reducing the span length (Murray, 1968; Levy, 1971; Peterson and Johnson, 1971; Baddeley *et al.*, 1984). No articulatory suppression is expected in a patient in which the rehearsal process is not operative.

Another way to distinguish patients with a deficit of the ph-STS from patients with a deficit of the rehearsal component is the different amplitude of the 'recency' effect (Waugh and Norman, 1965; Glanzer and Cunits, 1966) in free and in serial recall of sequences of verbal stimuli exceeding the length of the span. In normal subjects the free recall of verbal

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Fig. 1 MRI: T_1 -weighted image showing an isodense mass in the right cerebellar hemisphere (A); T_1 -weighted images showing the surgical excision of the posterior right cerebellar lobe (B and C).

stimuli assumes a U-shaped curve with the highest number of recalled items in the final positions of the list (recency effect) and, to a lesser extent, in the initial positions of the list (primacy effect). Since the recency effect is supposed to reflect the output of the ph-STS (Glanzer, 1972), a reduced recency effect is considered an expression of impaired ph-STS. Thus, in patients with reduced span but normal recency effect, the STM deficit can be reasonably attributed to a deficit in the rehearsal process (Vallar *et al.*, 1997).

Experimental tasks

The following experiments were performed in order to identify the functional locus of the patient's deficit within

	Feb. 5, '97	Feb. 18, '97	July 15, '97	Cut-off scores
Mini-Mental State	26/30*	29/30*	26/30*	23
Rey's 15 words				
Immediate recall	42.9	49.9	41.9	28.53
Delayed recall	8.2	9.2	9.2	4.69
Word recognition accuracy (%)	99	_	_	98
Digit span forwards	4*	4*	7*	7 ± 2
Digit span backwards	3*	3*	7*	5 ± 2
Spatial span forwards	4*	6*	-	7 ± 2
Spatial span backwards	4*	4*	-	5 ± 2
Immediate visual memory	-	17.2	-	13.85
Immediate picture recognition	97	100	-	91
Delayed picture recognition	94	100	_	94.2
Raven coloured matrices	24.8	24.8	-	18.96
Word fluency	11.4	23.4	23.4	17.35
Verbal analogies test	17	18	_	18
Temporal rules induction	39	40	_	15
Rey's complex figure (copy)	26.83	31.83	-	28
Rey's complex figure (recall)	14.69	20.19	-	6.20
Simple line cancellation	60/60*	60/60*	_	60
Double line cancellation	12/13*	12/13*	_	12
Wechsler adult intelligence scale				
Verbal IQ	89			
Non-verbal IQ	91			
Total IQ	89			

 Table 1 General neuropsychological examination: scores before/after surgery in February and July 1997

*Raw score: the other scores are adjusted, as explained in the Methods section.

Sequence length	Auditory-verbal		Auditory-pointing		Visual-verbal		Visual-pointing	
	Exam. 1	Exam. 2	Exam. 1	Exam. 2	Exam. 1	Exam. 2	Exam. 1	Exam. 2
1	1	1	1	1	1	1	1	1
2	1	1	1	1	1	1	1	1
3	1	1	1	1	1	1	1	1
4	0.8	1	1	1	0.6	1	1	1
5	0.2	0.6	0.6	1	0.4	0.6	0.4	0.2
6	_	0.6	0.2	0.2	_	0.2	_	0.2
7	_	0.2	_	_	_	_	_	_
Span	4	5.4	4.8	5.2	4	4.8	4.4	4.4

Table 2 Probability of correct sequences of digits by modality of stimulus presentation and response

Exam. 1 = Examination 1 (3 days after surgery); Exam. 2 = Examination 2 (5 months after surgery).

the STM system. The procedure adopted to explore the patient's STM deficit consisted of a shortened version of the procedure devised by Vallar *et al.* (1997). The original version of the test was modified in order to obtain a battery suitable for rapid and extensive assessment of a patient in a critical clinical condition whose deficit was expected to evolve quickly over time (Silveri *et al.*, 1997). The same tasks were administered 5 months later in order to document the evolution of the STM deficit. These were as follows.

Experiment 1. Digit span was assessed using various modes of presentation of lists of digits and various modes response.

Experiment 2. The rate of forgetting was assessed by

means of an Italian version (Vallar *et al.*, 1997) of the Brown–Peterson procedure (Brown, 1958; Peterson and Peterson, 1959).

Experiment 3. Phonological-similarity effects were assessed with lists of phonologically similar and dissimilar letters in auditory and visual presentations.

Experiment 4. The word-length effect was assessed with lists of words in auditory and visual presentations.

Experiment 5. The effect of articulatory suppression on the patient's immediate span was assessed using lists of visually presented digits.

Experiment 6. The recency effect was assessed by free recall of lists of words.

Experiment 1. Digit span

Five sequences of either one, two, three, four, five, six or seven digits were presented to the patient with a presentation rate of one digit per second. If the patient did not correctly recall at least one of the sequences the following longer list was not administered. This task was administered in four different conditions: (i) auditory–verbal (the patient repeats the sequence spelled out to him by the examiner); (ii) visual– verbal (the patient repeats the sequences of digits visually presented one-by-one by the examiner); (iii) auditory– pointing (the patient indicates the sequence of digits spelled out by the examiner from among alternatives printed on white cards randomly displayed on the table); and (iv) visual– pointing (the patient indicates the sequence of digits visually presented one-by-one by the examiner from among written alternatives).

Results. The results are summarized in Table 2. Auditory–verbal digit span was reduced in the patient (4) compared with normal subjects of the same age (Orsini *et al.*, 1987). The patient's digit span was significantly greater (5.4) four months later ($\chi^2 = 4.81$, P < 0.028).

(i) There was no significant advantage of the auditory modality over the visual modality, as is found in normal subjects (Conrad, 1964; Levy, 1971). However, a slight nonsignificant advantage was observed, but only when the response was made by pointing rather than verbally (auditoryverbal versus visual-verbal: 4 versus 4; and auditory-pointing versus visual-pointing: 4.8 versus 4.4). To some extent, this result contrasts with the observation that patients with STM deficit benefit from a visual presentation (for a discussion, see Shallice and Vallar, 1990) because of a reduced capacity of the ph-STS. In our patient, in the pointing modality, the slight improvement in the span results following the verbal presentation, compared with the visual presentation of stimuli, suggests at least a residual capacity of the ph-STS. An alternative explanation for this pattern of results, however, is that the phonological recoding of verbal stimuli in the visual presentation by the rehearsal process is defective. In any case, both interpretations concur in suggesting a deficit of the rehearsal rather than of the ph-STS. At the second examination 5 months later the usual advantage of the verbal presentation over the visual presentation, reported in normal subjects, was observed.

(ii) The patient showed a non-significant but consistent tendency to perform better when pointing rather than when the verbal response was requested, either with auditory (4.8 versus 4) or visual presentations (4.4 versus 4), suggesting a deficit at the level of the phonological output buffer component of the articulatory rehearsal. The performance in the auditory pointing condition (4.8) was within the normal range (range: 4.5–7) (see performances of normal control subjects in Vallar *et al.*, 1992). At the examination made five months later, both span modalities had improved (auditory–verbal span, 4 versus 5.4; auditory–pointing, 4.8 versus 5.2). However, only the increment in the verbal span was significant

 $(\chi^2 = 3.8, P = 0.05)$ confirming that, in the early stages, the verbal span was actually more impaired than the pointing span. Overall, the results converge in suggesting a deficit at the level of the rehearsal component of the STM, and in particular at the level of the phonological output buffer.

Experiment 2. Rate of forgetting

The patient was given the test described by Vallar *et al.* (1997) based on Brown and Peterson's procedure (Brown, 1958; Peterson and Peterson, 1959) in order to document the rapid forgetting of verbal material, a typical feature of patients with STM deficit. The patient was given 40 pairs of two-syllable words checked for word frequency. The patient's task was to repeat the two words of the pair in the same serial position of the presentation in four conditions: immediately after presentation (10 pairs), after 5 s (10 pairs), 10 s (10 pairs) or 15 s (10 pairs) of interpolated activity which consisted of counting backwards in twos from a two-digit number spoken aloud by the examiner.

Results. The results are described in Fig. 2. As typically occurs in subjects with STM deficit, the patient showed rapid forgetting; he forgot 50% of the words after 5 s of interpolated activity and 100% of the words after 15 s of interpolated activity.

In the second examination, the patient showed quite good recovery, since he forgot only 15% of the words after 15 s. The rate of forgetting was significantly lower in the second than in the first examination after 5 s ($\chi^2 = 7.62$, P < 0.058), 10 s ($\chi^2 = 5.23$, P < 0.02) and 15 s ($\chi^2 = 21.54$, P < 0.00005).

Experiment 3. Phonological-similarity effects

If, as suggested by the advantage of pointing over the verbal response, the STM deficit of the patient consisted of a reduced capacity of a component of the rehearsal system (the phonological output buffer), due to the impaired decoding of the visual stimuli into a phonological code, the phonological-similarity effect should be detected in the verbal but not in the visual presentation of stimuli. The patient was given five series of one, two, three, four, five and six phonologically similar (B, C, D, G, P, T, V) and phonologically dissimilar (F, K, Q, R, X, W, Z) letters (Baddeley, 1966) both in the verbal and visual modality of presentation. The response was verbal in both cases.

Results. The results are reported in Table 3. As expected, no phonological-similarity effect was detected with the visual presentation ($\chi^2 = 2.31$, P = 0.128) while the effect was confirmed with the verbal presentation ($\chi^2 = 4.11$, P < 0.0425). In the second examination the phonological-similarity effect was also significant in the visual presentation ($\chi^2 = 5.36$, P < 0.02).



Fig. 2 Rate of forgetting 3 days after surgery (February 1997) and 5 months later (July 1997).

Table 3 Probability of correct sequences of phonologically similar and dissimilar letters (phonological-similarity effect)

Sequence	Auditory-verbal		Visual-verbal (Exa	am. 1)	Visual-verbal (Exam. 2)	
	Similar	Dissimilar	Similar	Dissimilar	Similar	Dissimilar
1	1	1	1	1	1	1
2	1	1	1	1	1	1
3	0.4	1	1	1	1	1
4	0.4	0.4	0.8	0.8	0	1
5	_	0.4	0.2	0.4	0.6	0.8
Span	2.8	3.8	3	3.2	3.6	4.8

Exam. 1 = Examination 1 (3 days after surgery); Exam. 2 = Examination 2 (5 months after surgery).

Table 4 Probability of correct word sequences by word-length with auditory and visual presentations and verbal responses (word-length effect)

Sequence length	Auditory–Verbal (2 syllables)		Auditory–Verbal (4 syllables)		Visual–Verbal (2 syllables)		Visual–Verbal (4 syllables)	
	Exam. 1	Exam. 2	Exam. 1	Exam. 2	Exam. 1	Exam. 2	Exam. 1	Exam. 2
1	1	1	1	1	1	1	1	1
2	1	1	1	1	1	1	1	1
3	1	1	1	1	0.6	1	1	1
4	0.2	0.6	1	0.8	0	0.8	0	0.8
5	_	_	0	0.2	0	_	0	0.2
Span	3.2	3.6	4	4	2.6	3.8	3	4

Experiment 4. Word-length effects

As described above, in normal subjects, the immediate recall of lists of words is greater for short than for long words, due to efficiency of rehearsal. The patient was given five sequences of one, two, three, four and five, two-syllable and four-syllable words (see Vallar *et al.*, 1997) in auditory and visual presentation. If the rehearsal is impaired, no word-length effect should be observed in our patient.

Results. In the first examination there was a slight advantage

of the auditory presentation over the visual presentation (Table 4). This was in agreement with the observed tendency to obtain longer span with the auditory presentation. However, there was no significant effect of word length on the patient's recall. The advantage, if there was one, was unexpectedly in the opposite direction: an advantage with longer words. In the second examination, a general improvement in performance was confirmed. Once again, the patient did not show any significant effect of word length, and there was still a slight improvement with long words. The advantage



Fig. 3 Primacy and recency effects in the free recall of words; percentage of words correctly recalled, according to their serial positions (1st or 2nd, 3rd to 8th, and 9th or 10th).

of the auditory over the visual presentation was no longer present. The same task (in auditory presentation) was administered to two normal control subjects of the same age (18 years) and education (13 years). The first control subject showed a span of 4.8 for both short and long words; the second control subject had a span of 4.4 and 4.8 for short and long words, respectively. Thus, although the lack of word-length effect is taken as evidence of the inefficiency of the rehearsal process, we were unable to assign any specific value to this finding which would support a rehearsaldeficit hypothesis, as a similar result was obtained in normal subjects. The lack of a word-length effect, and even the existence of a paradoxical effect (better performance with longer words) in patients with impaired STM has also been described in other studies (patient TO of Vallar et al., 1997; Vallar and Cappa, 1987).

Experiment 5. Articulatory suppression

In subjects who rely on rehearsal of phonological information in the immediate memory span test, the continuous uttering of a nonsense syllable (such as 'blah') should disrupt the phonological information and produce a reduction of the span for both auditory and visual stimuli. In contrast, the interference should not occur if the rehearsal process is inoperative (as suspected in our patient).

In this case, as in other studies, the length of digit lists was based on the patient's performance level in recognition by pointing after visual presentation (four digits since the patient's performance was 4.4), and digit span was assessed with a pointing procedure (since the patient was engaged in uttering the nonsense syllables). An ABBA design was applied for the two experimental conditions, with and without (control) articulatory suppression. A total of 10 sequences of four digits were given in the articulatory suppression condition and a total of 10 sequences of four digits in the control condition.

Results. To some extent the result was unexpected, since the patient's performance was affected by articulatory suppression (5 correct sequences with articulatory suppression versus 9 correct sequences in control condition: $\chi^2 = 3.81$, P = 0.051) in spite of the supposed rehearsal impairment.

Experiment 6. The recency effect

A classical finding in patients with ph-STS store deficit is the reduced amplitude of the recency effect, i.e. of the ability to recall the items in the final positions of the list, in a free or serial recall of a series of stimuli that exceed the span. In order to explore the amplitude of the recency effect in our patient, five series of 10 words controlled for length and word frequency for free recall were administered. The task was to recall as many words as possible after each presentation. The amplitude of the recency effect is given by the total number of items recalled in the final positions of the list (positions 9 and 10)

Results. The results are shown in Fig. 3. In the first experiment, the patient recalled 4 out of 10 (40%) of the words in the final positions (positions 9 and 10) (recency effect) and 3 out of 10 (30%) of the words in the initial positions (positions 1 and 2) (primacy effect). Recall was lower in the intermediate positions (4 out of 30; 13%). In other words, there was no advantage of initial items over final items. The result ruled out any selective output deficit from the ph-STS (Glanzer, 1972; Vallar and Papagno, 1995), since it is consistent with a normal application of temporally based retrieval strategies to the content of the ph-STS. When the patient improved, a similar but higher-level U-shaped curve was obtained in free recall.

Discussion

The patient described here suffered from an STM deficit as demonstrated by his reduced digit span. The neuropsycho-

logical examination did not reveal other cognitive deficits. Before surgery word fluency and spatial span also seemed to be impaired but, a few days after surgery, these deficits were no longer present. In other words, the patient suffered from a selective impairment of the immediate retention of verbal information. This deficit was quickly resolved shortly and a few months later, performances on short-memory tasks were completely normal.

Since no cerebral lesion was detected in our patient in supratentorial structures and the only cerebral lesion was localized to the cerebellum, we can reasonably assume that there is some relationship between the cerebellar lesion and the reduced span. The STM impairment was transient. The rapid evolution of the patient's deficit does not minimize the relevance of our observation; on the contrary it confirms its causal relationship with the cerebellar damage. In fact, the observation of rapid cognitive recovery is typical in the clinical course of patients with acute cerebellar lesion (see, for example, the case of agrammatism or mutism following posterior fossa damage). On the other hand, a rapid recovery is also typical of the_movement disorders following cerebellar lesions and this does not challenge the role of the cerebellum in motor control.

Cognitive locus of this patient's STM deficit

The patient presented with a definite reduction of the immediate auditory verbal digit span when compared with control subjects matched for age (Orsini *et al.*, 1987); this is a typical finding in subjects with STM deficit. The patient also showed another typical feature of STM deficits, i.e. rapid forgetting of verbal stimuli, demonstrated by the Brown–Peterson paradigm. Immediate repetition of individual items proved to be intact, ruling out the hypothesis of more peripheral language deficits due to impaired decoding of verbal stimuli or of impaired articulatory realization of verbal output.

The first way to identify the locus of the patient's deficit within the STM system lies in the interpretation of the results obtained in the various span conditions. The patient's span tended to be better when the responses were given by pointing rather than verbally. The advantage of pointing over the verbal response in this patient suggests some impairment at the level of the phonological output buffer (Vallar et al., 1997) component of the rehearsal process of the STM system. His performance in the auditory pointing condition (4.8) in the first examination after surgery was still within the normal range defined on the basis of the performances of 12 normal control subjects reported in Vallar et al. (1992) (range 4.5–7), even though slightly less than his performance at the second examination. A deficit of the rehearsal locus was also suggested by the advantage of pointing after the verbal presentation of stimuli compared with the visual presentation. In fact this result suggests either a residual capacity of the STS or a deficit of the phonological recoding of visual

stimuli, a recoding which relies on the rehearsal component of the STM system.

The confirmation of a deficit at the level of the rehearsal component derives from the fact that the patient presented phonological-similarity effects with auditory, but not with visual, presentation of letters. In other words, with visual modality of presentation his span for phonologically dissimilar letters was not significantly greater than the span for phonologically similar letters, suggesting reduced efficiency of the rehearsal process in the phonological recoding of visual stimuli.

There is no easy interpretation of the results obtained with the word-length paradigm. No word-length effect was observed, either for auditory or for visual presentation of the words. The advantage conferred was actually observed in the opposite direction to that expected, i.e. the span was longer for long words than for short words. The lack of the wordlength effect in patients with rehearsal deficits or similar paradoxical effects has already been described (Vallar and Cappa, 1987; patient TO of Vallar *et al.*, 1997). Interestingly, the advantage for the span of long words over short words was confirmed when our patient's STM deficit improved, and similar results were obtained from two matched normal control subjects.

One possible interpretation of the paradoxical effect may lie in the discussion of the 'trace decay plus rehearsal' theory (Stigler et al., 1986; Baddeley, 1990; Schweickert et al., 1990) offered by Brown and Hulme (1995). According to this hypothesis, the recall of an item in a span-measurement task involves reconstruction of the item from a partially decayed phonological trace. This process may be length sensitive. If just a phonological segment is lost, long items are expected to be easier to reconstruct than shorter items for which correct recall is less probable. This and other factors of a methodological order might account for the inconsistent, and even paradoxical, results obtained by various authors in the word-length paradigm. Thus, we are unable to interpret our results as supporting the rehearsal deficit. In support of such a deficit, we can only emphasize the better performance in the auditory-verbal modality compared with the visual-verbal modality, confirming our patient's difficulty in recoding visual information.

Evidence in support of a rehearsal locus for our patient's STM deficit can be found in the presence of a U-shaped curve in free recall, i.e. a normal amplitude of the recency effect. The amplitude of the recency effect reflects output from the ph-STS (Glanzer, 1972; Vallar and Papagno, 1995); thus, a normal recency effect is considered to be an expression of a normal ph-STS and the reduced immediate retention of verbal memory is traced back to an inefficient rehearsal process.

Special attention should be given to the observation that in our patient articulatory suppression reduced the span, in spite of the hypothesized deficit of the rehearsal process. A plausible explanation for this unexpected result may be advanced. As stated previously, the articulatory suppression is supposed to inhibit the rehearsal process which, as already mentioned, makes use of the phonological output buffer. The procedure adopted to explore the articulatory suppression effect bypasses the phonological output buffer; in fact, since the patient is engaged in the articulation of nonsense syllables, his span is necessarily evaluated by means of a pointing procedure. In other words, the procedure adopted excludes precisely the component of the rehearsal process supposed to be impaired in our patient. We demonstrated, in fact, that digit span was close to normal when a pointing procedure for responding was requested.

To conclude, the hypothesis is that our patient suffers from a disproportionate impairment of the phonological output buffer component of the rehearsal system, with a preservation of the ph-STS component. This cognitive locus of the deficit is consistent with the pattern of results obtained: the improvement of the span with the pointing procedure rather than the verbal response; the advantage of auditory presentation over visual presentation of stimuli; the lack of phonological-similarity effect with visual presentation of stimuli. On the other hand, the functioning of the ph-STS is demonstrated by the normal amplitude of the recency effect and by the phonological-similarity effect with auditory presentation.

The cerebellum and STM deficits

Activation of the cerebellum during tasks engaging verbal STM has been demonstrated in PET studies (Paulesu et al., 1993; Awh et al., 1996; Fiez et al., 1996; Paulesu et al., 1996). Paulesu et al. (1993) demonstrated that the two subsystems of the STM have discrete neural correlates. In their study they measured cerebral blood flow while subjects were performing two different tasks: the first (short-term memory for letters) was supposed to involve both components (ph-STS and rehearsal) of the STM system; the second (rhyming judgements on visually presented letters) involved only the rehearsal process. Comparison of distribution of activity in these two conditions localized the ph-STS in the left supramarginal area and the subvocal rehearsal component in Broca's area. Interestingly, in their study, the subvocal rehearsal component also activated Broca's area, the supplementary motor area, the insula and right cerebellum while the short-term memory for letters, which is supposed to rely on the ph-STS but not on rehearsal, involved only the left supramarginal area. This suggests that the cerebellum belongs to a complex system that subsumes some aspects of language planning and execution, in particular the silent rehearsal of verbal material. This system involves the left inferior frontal gyrus (Brodmann area 44 or Broca's area), the left supplementary motor area (area 6) and the insula. Direct neuropathological evidence of the relationship between damage of specific supratentorial neural substrates and differential impairment of the two components of the ph-STS-rehearsal system has been provided by Vallar et al. (1997) who described two patients with differential impairment of the two components of the STM system. One of the patients, with

reduced span due to impairment of the ph-STS, had a lesion extending to the left inferior parietal lobe, and superior and middle temporal gyri. The other patient, who had reduced capacity of the phonological output buffer, had a left hemisphere lesion involving the premotor and the rolandic regions, the frontal paraventricular area and the anterior part of the insula. Thus, there is high consistency between functional studies and neuropathological data concerning the supratentorial areas underlying the ph-STM store-rehearsal system. Our finding confirms the functional studies with regard to the cerebellum, providing direct evidence that a cerebellar lesion may be related to a deficit of the rehearsal component, in particular of the phonological output buffer.

In a model in which they try to combine aspects of the functional architecture of models of single word production and immediate verbal memory, Burani et al. (1991) conceive the phonological output buffer as a working memory space in which phonological segments are temporarily stored prior to the application of the various output processes, such as planning and editing the articulatory procedures needed to produce speech. This component would also be involved in two other functions, i.e. in the rehearsal of information held in the ph-STS, and in conveying visually presented information to the ph-STS. In other words, the phonological output buffer component would be involved both in the articulatory programming of speech output and in the silent rehearsal process. However, the hypothesis (Klapp et al., 1981), that the articulatory programming of speech output (phonological output buffer) and the rehearsal component may be distinct processes, is not definitely excluded (for a discussion of this issue, see also Shallice and Vallar, 1990). The pattern of results obtained in our patient would seem consistent with this possibility; in fact, the patient's implicit speech rehearsal proved to be still operative (the articulatory suppression reduced the span) in the presence of defective articulatory programming of speech output. Silent rehearsal of verbal material and articulatory planning of speech might utilize different neural structures. While the rehearsal might be a function of supratentorial structures, the articulatory planning might be a function of the cerebellum. In other words, the cerebellum might operate a control of speech production at the interface between the subvocal rehearsal process and the motor execution. A deficit of the articulatory programming due to cerebellar lesion should still be considered a 'high level' deficit, since it has been demonstrated that articulatory planning in the phonological output buffer does not require any implementation of articulatory codes from the peripheral musculature (Baddeley and Wilson, 1985; Vallar and Cappa, 1987). This conclusion should be emphasized since it is consistent with the fact that our patient did not present any overt articulatory disorder in speech output.

The cerebellum and speech production

There is long-standing clinical evidence that a cerebellar lesion may produce disorders in articulation of speech (Amarenco *et al.*, 1991; Ackermann *et al.*, 1992; Barth *et al.*, 1993). A more general relationship between speech production and the cerebellum is also suggested by the development of transient mutism after resection of cerebellar tumours in children (Pollack, 1997) and adults (Dunwoody *et al.*, 1997). The above-mentioned data from functional studies suggest that the cerebellum is also involved in covert articulation (for a discussion, see Fiez and Raichle, 1997). Our study supports this view, offering direct evidence that a cerebellar lesion may affect the 'silent articulation' of verbal information, i.e. the articulatory program of speech output within the phonological output buffer.

In cerebellar patients, disorders of overt articulation seem to be correlated with left cerebellar lesions (Lechtenberg and Gilman, 1978; Amarenco et al., 1991). Functional studies demonstrate cross cerebellar diaschisis after frontoparietal infarct (Pantano et al., 1986), confirming anatomical data on the existence of crossed cerebrocerebellar connections (Allen and Tsukahara, 1974). The hypothesis has been advanced that severe dysarthria observed after right hemisphere damage might be interpreted on the basis of such a connection between the right hemisphere and the left cerebellum (Lechtenberg and Gilman, 1978; Ropper, 1987). Our results suggest that the cerebellum is also involved in covert recirculation of verbal information. This function might be underpinned by the crossed connections between the right cerebellum and the anterior regions of the left hemisphere, where damage has been demonstrated in patients with impaired rehearsal of verbal information in the STM system. Finally, the clinical and experimental evidence available until now suggests that the cerebellum takes part in both aspects of speech production: in overt articulation by means of a functional system involving the left cerebellum and the right hemisphere and in covert articulation by means of a functional system involving the right cerebellum and the anterior left hemisphere regions. Within this view, transient mutism observed after cerebellar excision might be attributed to a global involvement of both the articulatory functions (covert and overt) of the cerebellum. Although the exact neurobiological substrate of post-operative mutism has not yet been elucidated it has been hypothesized that this deficit may result from injury to the afferent and/or efferent projections that pass through the dentate or interposed nuclei bilaterally (Pollack, 1997). Since the complex reciprocal connections between the cerebellum and cerebral hemispheres traverse these nuclei (Schmahmann, 1996) the involvement of both overt and covert articulation is plausible. However, very little neuropsychological evidence is available on the nature of the cognitive deficit in subjects who developed mutism after cerebellar surgery, and specific studies on this issue are necessary to confirm this hypothesis

Our results may also contribute towards a better understanding of the agrammatic speech reported in patients with right cerebellar lesions (Silveri *et al.*, 1994; Zettin *et al.*, 1995). The hypothesis advanced by Silveri *et al.* (1994) is that the reduced capacity of the phonological output buffer impairs the retention of the phonological/morphological

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components as long as necessary for the syntactical implementation of the phrase. It might be argued that our patient is not agrammatic in spite of the reduced capacity of his phonological output buffer. His only deficit in the linguistic domain was, in fact, a transient reduced verbal fluency. Similarly, it has been argued that patients with obvious limitations in immediate verbal span may not actually present the supposed STM-related disorders (e.g. sentence comprehension deficit and repetition disorders). However, it may be that different STM-related deficits lie on a continuum of STM impairment severity; see, for example, the discussion by Martin (1996) on a STM-based account of repetition disorders within an interactive activation model. In addition, different deficits may be produced by the differential impairment of the various components of the STM system. In other words, quantitative/qualitative differences in STM impairment could account for the range of syndromes that may be potentially found in association with a reduced verbal span, for example, with regard to the cerebellar lesions, a (transient) reduced verbal fluency, as in our patient (see also Leggio et al., 1995), or agrammatism, as in patients described by Silveri et al. (1994) and Zettin et al. (1995). Further clinical and experimental evidence is necessary for a better definition of the role of the cerebellum in the various steps of speech production.

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