Neurolinguistic and follow-up study of an unusual pattern of recovery from bilingual subcortical aphasia

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Summary

We report on the neuropsychological and neurolinguistic features of a bilingual patient, E.M., who presented with an uncommon pattern of aphasic deficit consequent to subcortical lesions mainly involving the left basal ganglia. Not only are reports of bilingual subcortical aphasia rare, but E.M.'s deficit is particularly uncommon for it concerns the most used mother tongue (Venetian) much more than a less practiced second language (standard Italian). In this patient, the linguistic deficit in mother tongue production has been observed in spontaneous speech and in cross language translation tasks, where an asymmetrical paradoxical performance has been revealed. Indeed, unlike neurologically intact subjects, E.M. has more difficulties when translating into her mother tongue than into her second language. Although E.M.'s mother tongue is prevalently an oral language, the asymmetrical translation pattern is similar in written and oral translation tasks, thus ruling out the possibility that the deficit simply reflects differences between written and oral language codes. Finally, another remarkable feature of E.M.'s impairment is its stability over almost 5 years from the stroke. We propose that this unusual type of recovery in E.M. is related to the higher degree of automatization of the first language with respect to the second one. This proposal fits with the role of basal ganglia in automatized motor and cognitive performance.

Keywords: bilingualism; subcortical aphasia; basal ganglia; implicit and explicit memory

Abbreviations: L1 = mother tongue (Venetian); L2 = second language (standard Italian)

Introduction

Although several psychological and linguistic markers have been used to characterize bilinguals and polyglots (Albert and Obler, 1978; Grosjean, 1982; Hamers and Blanc, 1989), the best criterion for classifying these subjects is still a pragmatic one. People who speak and understand two languages, or two dialects, or one language and one dialect, and who are able to avoid mixing the two linguistic systems when writing or speaking, can be referred to as bilinguals (Fabbro, 1996). There are at least two reasons why a subject who speaks and understands both a language (e.g. standard Italian) and a dialect (e.g. Calabrian) can be considered bilingual. First, it is impossible to classify by means of unequivocal criteria concerning whether or not the language spoken by the members of a given community is a dialect or a language (Tagliavini, 1969; Hagège, 1985; Pinker, 1994). The so-called Chinese dialects, for example, differ from each other in terms of linguistic cognateness much more than many standard languages spoken across Europe (Chomsky, 1977). Secondly, it is not known whether cognate languages (e.g. Spanish–Catalan), or unrelated languages (e.g. English–Japanese), or dialects are separately represented in the bilingual brain (Paradis, 1989, 1993).

Although there is some controversy on the functional organization of the bilingual brain, relevant information on this issue is available from patients with aphasic deficits consequent to stroke (Ojemann and Whitaker, 1978; Paradis, 1989). The analysis of recovery patterns in bilingual aphasics shows that the different languages often recover over the same time course and to a similar extent (parallel recovery). It is also common that recovery is better or even selective for the mother tongue or the most familiar language at the time of the stroke (Paradis, 1977; Fabbro, 1996). However, the analysis of less standard recovery patterns is potentially highly informative (Paradis, 1977, 1989, 1993; Albert and Obler, 1978; Fabbro, 1996). For example, a type of bilingual aphasia where the patient's performance in one language improves while performance in another language deteriorates (antagonistic recovery) has been described. A variant of this pattern is the so-called alternate antagonism in which patients seem to have access to only one of their languages for alternating periods of time, e.g. a day or a week (Paradis, 1989). Another non-standard recovery is the so-called differential aphasia, in which patients show, for example, a Broca's aphasia in one language and a Wernicke's aphasia in another (Albert and Obler, 1978; Silverberg and Gordon, 1979). Such a pattern would provide support for distinct and largely separate neural bases for the different languages. It is worth noting, however, that a given kind of aphasia may induce different symptoms across the different languages, e.g. patients affected by Broca's aphasia may omit freestanding grammatical morphemes in English, but add or substitute bound grammatical morphemes in Italian (Miceli *et al.*, 1989). On this basis, it has been claimed that there is no proof for the existence of true differential aphasia (Paradis, 1995).

Several factors may play a role in the non-standard patterns of language recovery in bilinguals and polyglots. It has been reported that patients may preferentially recover the language spoken in the hospital soon after their stroke (Bychowsky, 1919), or the language used under particular circumstances, e.g. at school (Kainz, 1960) or for praying (Kraetschmer, 1982). Halpern (1941) reported a patient in whom reading and writing brought about a selective recovery, thus suggesting that languages based on both oral and written codes may be more resilient to aphasic disruption than languages relying only upon oral codes. More recently, Paradis (1994) postulated that the mother tongue may rely upon memory systems which differ from those used for other languages, especially when the latter are acquired later in life. It has been suggested, in fact, that the mother tongue is learned and used implicitly, i.e. according to automatic rules that are largely impervious to consciousness (Ellis, 1994). By contrast, a second language, particularly if learned in adult life, is probably learned and used explicitly, i.e. mainly by consciously applying rules. Moreover, clinical and neuroimaging studies suggest that implicit and explicit memory systems do rely upon different neural structures (Saint Cyr *et al.*, 1988; Perani *et al.*, 1993; Shimamura, 1993; Ostergaard and Jernigan, 1993; Butters *et al.*, 1994; Schacter, 1995; Squire and Knowlton, 1995). In particular, while implicit memory heavily relies upon subcortical structures, like the basal ganglia and the cerebellum, explicit memory relies mainly on a widely distributed cortical network.

Aphasia has been classically considered a consequence of cortical lesions. However, neuroimaging techniques in monolinguals have revealed that aphasia may also occur after lesions confined to the thalamus, the basal ganglia or the paraventricular white matter (Alexander, 1989). In particular, monolinguals who become aphasic after lesions involving the basal ganglia may present with (i) a reduction of spontaneous speech, (ii) paraphasic errors both verbal (e.g. patients say daisy for table) and semantic (e.g. patients say spoon for knife), (iii) neologisms, perseverations and echolalias or (iv) spared or only mildly impaired comprehension and repetition (Wallesch and Papagno, 1988; Alexander, 1989; Démonet *et al.*, 1991; Crosson, 1992). While subcortical aphasia in monolinguals is rather well known, only a few cases of bilingual subcortical aphasia have been reported. Moreover, although there is some PET evidence suggesting the involvement of the left putamen in bilinguals' speech production (Klein *et al.*, 1994, 1995), no accurate clinical and radiological studies of bilingual aphasic patients with lesions to the basal ganglia have hitherto been reported.

The patient (E.M.) described in the present paper is the first known case of bilingual aphasia in which the lesion is confined to subcortical structures and mainly involves the left basal ganglia. Also remarkable is the fact that E.M.'s linguistic deficit mainly involves her mother tongue and that her second language, although less practiced, is relatively spared. The present study is an extension of a previous report by Aglioti and Fabbro (1993).

Case report

Clinical history

Patient E.M., a right-handed housewife, was born in March 1922. In November 1990, she suddenly suffered from a mild right sensorimotor hemisyndrome with a 2-week period of mutism. According to medical records and the description delivered by the patient herself and her relatives, the main residual impairments in the first few months after the stroke consisted in a slowing down of movements and a remarkable speech deficit characterized by the fact that E.M. was no longer able to speak Venetan (Veronese dialect). It is relevant to note here that linguistic geography in Italy is rather complex. In addition to a large number of regional dialects (Venetian, Piedmontese, Ligurian, Neapolitan–Campanian, Sicilian, Calabrian, etc.) and two so-called minority languages (Friulian and Sardinian), several foreign languages (German, French, Slovenian, Albanian and Catalan) are spoken across the country (Salvi, 1975). Most of the dialects are so different from standard Italian that a person speaking in a given dialect cannot be understood by another Italian who has never been exposed to it. In addition, there are several dialect variants in each region. In particular, Venetan is a Romance language spoken in north-eastern Italy. It includes the dialects spoken in Venice (Venetian), Verona (Veronese), Treviso (Trevisan) and Padua (Paduan), which are predominantly oral dialects (Tagliavini, 1969). The differences between Venetan dialects and standard Italian mainly concern phonological and intonational aspects. Relevant differences on the lexical (Appendix 1) and syntactic level (Appendix 2) are also present.

Venetan was E.M.'s mother tongue and the language she had been using all her life; nonetheless, she presented with a very strong tendency to use standard Italian even when her relatives and the medical staff addressed her in Venetan. During the first 3 months following the brain insult, the patient's mother tongue was so severely impaired that she could hardly interact linguistically with her family and friends. Eleven months after the stroke the patient spontaneously applied to the speech therapy service at the Ospedale Policlinico in Verona, asking to be re-educated in the comprehension and production of her mother tongue. Both E.M. and her relatives found the nature of the linguistic deficit extremely odd and they had not realized that E.M. had such a high proficiency in speaking standard Italian.

Neurological examination

In November 1991, the first time we gave E.M. a neurological examination, the patient showed a slight tendon hyperreflexia of the right upper limb, a slight deficit in fine digital movements on the right side and a general postural instability. Her rate of speech and movements appeared slow. No problems with cranial nerves were detected.

Neuroradiological findings

A CT scan exam performed 1 week after the stroke showed hypodensity of the left capsulo-putaminal region probably consequent to ischaemia. An MRI scan, performed in April 1992, i.e. ~16 months after the stroke, provided more detailed information about the extent of the lesion (Fig. 1A and B).

Neuropsychological findings

In November 1991, when E.M. was given a formal neuropsychological examination, she was perfectly oriented in space and time. Her intelligence, as inferred from the Wechsler Adult Intelligence Scale (administered in standard Italian) was within normal range considering her age and her low level of education (*see* below). A detailed profile of E.M.'s performance in the different sub-tests is reported in Table 1.

Table 1 Patient E.M.'s scores on the Wechsler scale

	Wechsler Scores	
	E.M.'s score	Controls (mean±SD)
Information	11	9.5±3.2
Comprehension	13	8.9±2.3
Arithmetic	4	9.5±3.6
Similarities	10	8.8±2.8
Digit span	5	8.8±2.9
Vocabulary	12	9.6±3.2
Digit symbol	8	5.4±2.8
Picture completion	9	7.1±2.3
Block design	6	7.8±2.8
Picture arrangement	12	6.9±2.5
Object assembly	7	7.8±2.9

The control data is from 206 non-brain-damaged, age-matched controls.

Her short-term spatial memory examined by using Corsi's block tapping test was within the normal range. She scored four blocks (reference values 4.3±0.8, Spinnler and Tognoni, 1987). Also in the learning of a supraspan sequence of Corsi's blocks to be reproduced twice in a row without errors, she scored 11.6 (within the normal range). Reference values obtained from a sample of 48 non-brain-damaged individuals matched for age and education were 16.5±7.1 (Spinnler and Tognoni, 1987). Ideomotor gestural apraxia was tested and scored according to De Renzi *et al.* (1980). Patient E.M. turned out to be nonapraxic, scoring 61 out of 72 with the right hand and 64 out of 72 with left hand (cut-off score = 53) and no impairment with the use of objects was detected. She showed oral apraxia by scoring only 16 correct responses in a standard test where 48 age matched controls scored on average 19.79±0.58 SD (Spinnler and Tognoni, 1987). She was given the Wisconsin Card Sorting Test, a task based on the ability to switch between different sorting principles (Milner, 1963), for the purpose of assessing the possible presence of frontal deficits. The patient reached the maximum possible number of sorting criteria (six times using 125 cards) and made 17 perseverative errors and 20 non-perseverative errors. These numbers of errors were lower than those observed in a large sample of frontal and non-frontal brain damaged patients (Anderson *et al.*, 1991). Patient E.M. appeared hypomimic, but she was not poor in recognizing emotional facial expressions. She showed high proficiency in a task requiring judgment of verbal intonation (as in interrogative sentences). She was able to detect the calabrian regional accent of one examiner (S.A.) who always addressed her in Italian. Moreover, E.M. was quite able to report that one of the examiners (F.F.) addressed her in Friulian.

Linguistic deficits

From a series of interviews, some of which specifically regarding the patient's story of bilingualism (Paradis, 1987), it has been established that E.M. grew up, and had always lived, in a family environment where only her mother tongue, Venetan (L1) was spoken; her formal education consisted of only 3 years of elementary school in her second language, Italian (L2). The patient usually watched Italian TV and read Italian magazines, but she hardly ever spoke in her second language (two or three times in a year) and then only with great effort and using several words from her L1. Thus, it appeared rather odd that during preliminary informal conversations E.M. spontaneously spoke L2 only, even when specifically addressed in L1. Her speech production was non-fluent, slow and characterized by a low voice. She hardly ever produced spontaneous speech in her mother tongue and on request she could, with a great effort, produce only a few words. Moreover, she spoke L2 with a foreign accent, a symptom which is related to lesions of the left basal ganglia (Graff-Radford *et al.*, 1986; Blumstein *et al.*, 1987; Gurd *et al.*, 1988; Alexander, 1989).

Patient E.M. was administered the Italian version of the Aachener Aphasie Test (Luzzatti *et al.*, 1991). In addition, an *ad hoc* translation in Venetian of the Aachener Aphasie Test test was also given to the patient. Although she claimed to be unable to understand L1 during the first 3 months after the stroke, her performance in the two versions of the Aachener Aphasie Test test showed that, while comprehension was similarly affected in the two languages (e.g. in the Token test L1 = 35 out of 50 and L2 = 36 out of 50), language production was selectively impaired in L1. In oral naming tasks, e.g., the incidence of correct responses was 63 out of 120 in L1 and 105 out of 120 in L2 [($\chi^2(1)$ = 33.35, $P < 0.0001$]. In addition, in an object naming task E.M. scored 29 out of 30 in L2 and 12 out of 24 in L1 [$\chi^2(1)$ = 15. 88, $P < 0.0001$]. When she was asked to speak in L1, errors consisted of four semantic paraphasias, seven correct words uttered in L2 and one anomia. In the description of pictures she scored 28 out of 30 in L2 and 3 out of 30 in L1 [$\chi^2(1) = 41.71, P < 0.0001$]. Thus, E.M.'s condition appeared similar to a Broca's aphasia in both L1 and L2.

The rather uncommon dissociation observed in E.M., between L1 and L2 in language production, was systematically evaluated by analysing spontaneous speech performance in both languages, oral translation of words and sentences from L1 into L2 and vice versa, and a written cross-language translation of words. The experimental tests were delivered twice per week, in sessions of ~60 min each, during the period from November 1991 to May 1992. There was one follow-up session of ~3 h was carried out on July 25, 1995 (*see* below). Informed consent was obtained after the non-therapeutic nature of the study was explained to the patient and her relatives. All tests were administered by a Venetan/Italian bilingual speech therapist. Since all testing sessions were video-recorded, results were scored with the aid of film sequences.

Experimental tasks

Spontaneous speech in L1 and L2

In each of the spontaneous speech sessions only one of the languages was used all the time the examiner and the patient were together. Spontaneous speech analysis was conducted according to Paradis (1987). Results are summarized in the left part of Table 2. It is important to note that during sessions

(A)

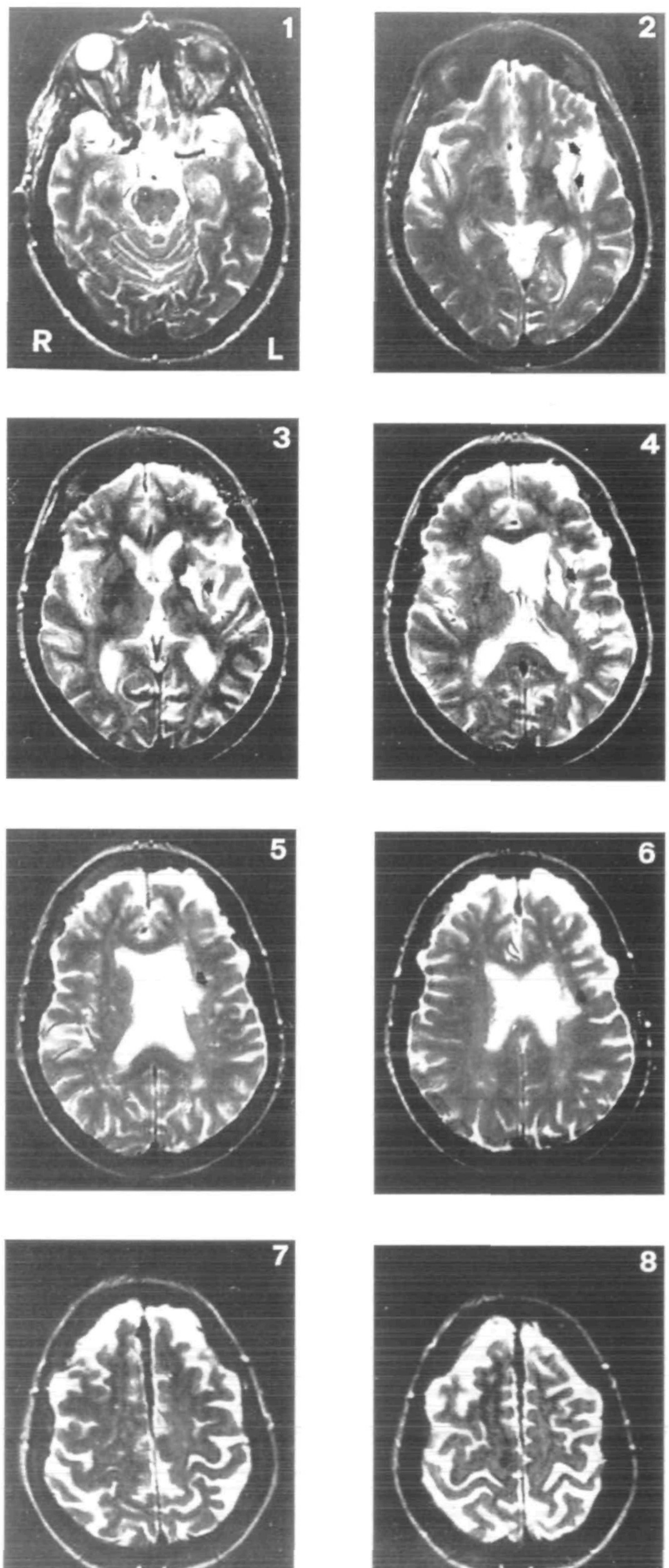

Fig. 1 Transverse (**A**) and coronal (**B**), T_2 weighted MRI scans. The number on the upper right corner of each scan marks the caudal to rostral progression of the transverse slices, and the anterior to posterior progression of the coronal slices. The main lesion, indicated by the black arrows, appears white. A slight cortical atrophy is apparent. Small lesions of the subcortical white matter are present bilaterally.

in L1 there was a high percentage of words (51.7%) in the non-requested language (L2); by contrast, the percentage of words in L1 during L2 sessions was rather low (4.4%). This result is even more striking considering that during the sessions where only Venetan was allowed the patient was reminded several times to speak Venetan, because she violated instructions by inadvertently switching to Italian. In contrast, during sessions in L2 no reminders were necessary. Patient E.M. showed a low verbal fluency in both L1 (72.8 words per minute and 4.9 words per phrase) and L2 (78.4 words per minute and 5.1 words per phrase). Note that a sample of controls, asked to produce spontaneous speech in standard Italian, uttered ~130 words per minute with 10 words per phrase (Miceli and Mazzucchi, 1990). Patient E.M. also omitted ~10% of grammatical morphemes in both L1 and L2. In addition to the above-reported signs of non-fluent aphasia, she also showed signs of fluent aphasia and substituted and added free-standing grammatical morphemes in inappropriate contexts. Also, she produced neologisms, and verbal and semantic paraphasias (Table 2). Moreover, there were perseverations and echolalic repetitions of words in both L1 and L2. During L1 sessions, E.M. produced 11 utterances which were entirely echolalic. Four utterances (36%) were spontaneously translated into L2.

Cross-language translation tasks

Patient E.M. was asked to translate from L1 into L2 (and vice versa) separate lists of words and sentences. All words and sentences administered in these tasks had been previously chosen from the Veronese–Italian dictionary (Beltramini and Donati, 1982). When choosing the words and sentences for

(B)

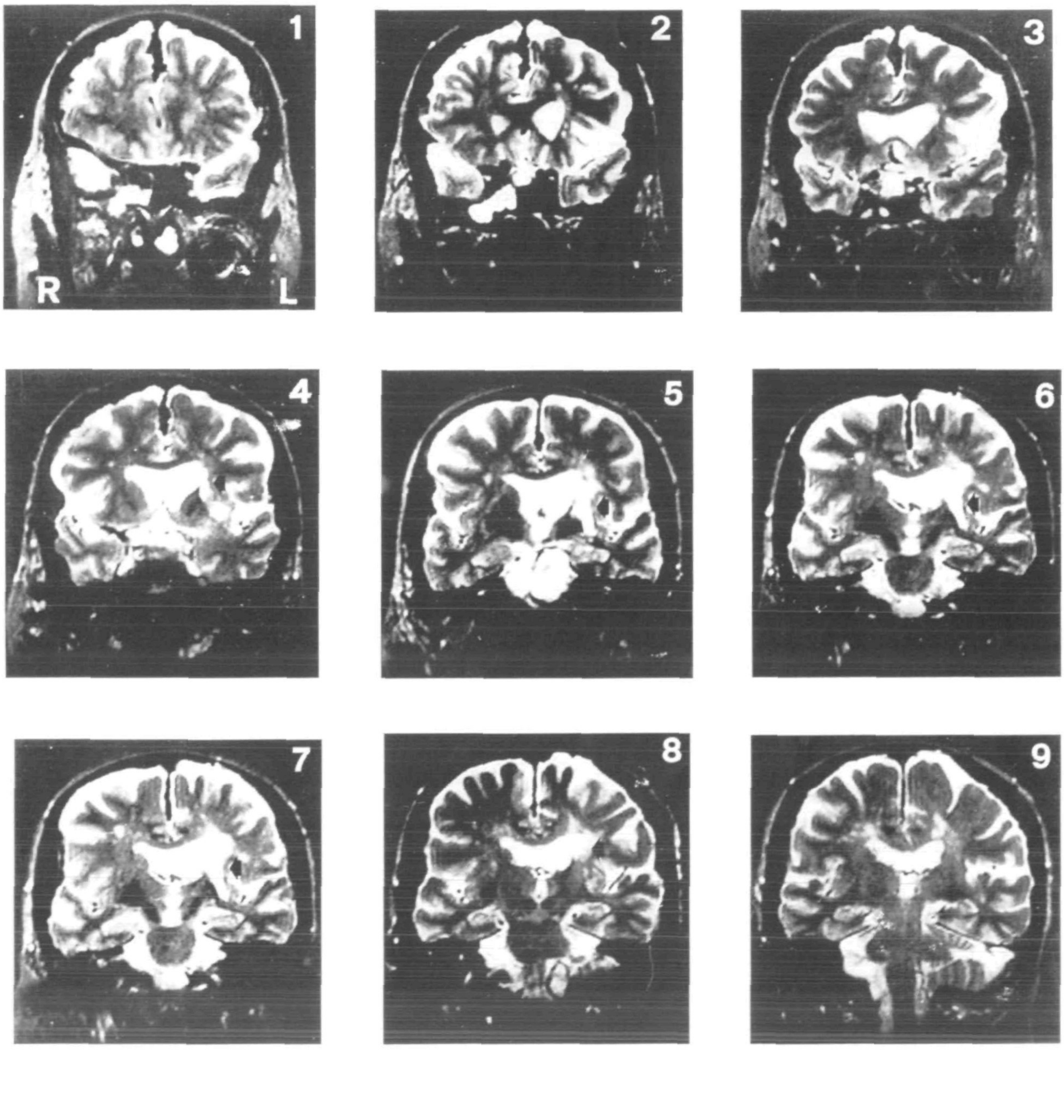

Table 2 Analysis of Patient E.M.'s spontaneous speech in L1 (Venetan) and L2 (Italian)

	Spontaneous speech after the lesion			
	One year		Five years	
	L1	L2	L1	L2
Tape recording time (minutes)	45	33	2.83	1.75
Time during which E.M. spoke (minutes)	23.5	16	2.17	1.5
Total number of words	1710	1245	113	106
Words per minute	72.7	78.4	52.3	70.7
Number of utterances*	346 (20.2)	224 (17.8)	19 (16.8)	21 (19.8)
Word-finding difficulties	64 (3.7)	37 (2.9)	4 (3.5)	2 (1.8)
Agrammaticisms	100 (5.8)	101 (8)	4 (3.5)	3 (2.8)
Paragrammaticisms	21 (1.2)	33 (2.6)	1 (0.8)	0
Niologisms	15 (0.9)	9 (0.7)	0	0
Phonemic paraphasias	39 (2.3)	25 (2)	2 (1.8)	0
Semantic paraphasias	6 (0.3)	1 (0.1)	0	0
Verbal paraphasias	3 (0.2)	6 (0.5)	0	0
Echolalic words	24 (1.4)	14 (1.1)	2 (1.7)	0
Perseverations	100 (5.8)	46 (3.7)	10 (8.8)	1 (1.1)
Words in the other language	885 (51.7)	55 (4.4)	62 (54.8)	3 (2.8)

The % ratio of the number of utterances to the total number of words is shown in parentheses. For each type of error, values in parentheses express (in %) the ratio of the errors to the total number of words uttered by the patient. Agrammatisms refer to omissions of free-standing grammatical morphemes. Paragrammatisms indicate substitution or addition of grammatical morphemes (Miceli *et al.*, 1989). Phonemic paraphasias refer to the number of words containing phonemic errors. Words uttered by the examiner which were immediately repeated by E.M. are referred to as echolalic. Perseverations indicate inappropriate, immediate, successive repetition of words. The amount of time spent speaking in the two languages mainly reflects two factors: (i) E.M. was slower in producing L1; (ii) for rehabilitation purposes, sessions dedicated to speech in L1 were more numerous than sessions dedicated to speech in L2. *According to Paradis (1987) an utterance is defined as a self-contained segment of speech conveying its own independent meaning. Although a sentence is an utterance, an utterance may be shorter than a complete sentence (Paradis, 1987).

the translation tests, two main criteria were followed: (i) only words and sentences with exactly corresponding meaning in the two languages but different lexical stems and syntactic structures were chosen; (ii) only common words were used. Words and sentences used in the different translation tasks are reported in Appendix 1 and 2, respectively. In both appendices, each item is reported first in standard Italian and then in Venetian. The words in the list could belong to four grammatical classes, namely verbs (19), adverbs (5), concrete nouns (41) and abstract nouns (10). The same lists of words and sentences were used in both directions of translation. To avoid interference phenomena, each session was devoted to only one direction of translation (e.g. from L1 into L2). To avoid biases in favour of E.M.'s linguistic asymmetry, translation from L1 into L2 was performed before the opposite condition. By adopting this test order, we might have induced a bias towards a better translation into L1. Consequently, any translation pattern overcoming such bias must be considered a very robust result. E.M.'s husband (A.B.), a 69-year-old, right-handed man, with 4 years of schooling in L2 served as a control in the translation tasks.

Word translation tasks

A quantitative analysis of E.M.'s and A.B.'s performances in the word translation tasks is reported in Fig. 2. Errors were also analysed from a qualitative point of view. When asked to translate 75 words from L2 into L1, E.M. simply repeated nine words without translating them; in three cases she produced a calque [e.g. the word 'trovare' (to find) was translated as 'trovar' instead of 'catàr']; in two cases she produced synonyms in the source language [e.g. the word 'somaro' (donkey) was translated as 'asino' (ass), instead of 'mùsso']; she also produced four circumlocutions, one semantic paraphasia and three neologisms in L2; finally, in 22 cases she simply gave no response (or reported she did not know the word). When asked to translate 75 words from L1 into L2, E.M. made 11 circumlocutions in L2, two semantic paraphasias in L2 [e.g. she translated 'sgaùia' (garbage) as 'scopa' (brush)]; in five cases she uttered correct L2 words which had nothing in common with the source-language words, and in five cases she gave no response. The asymmetry in translation did not selectively affect any stimulus category, being evenly distributed across verbs, adverbs, and concrete and abstract nouns.

The only two errors made by the control subject during translation into L1 were one failure to respond and one semantic paraphasia ['spossatezza' (tiredness) was translated as 'stràco' (tired) instead of 'lèla']; five out of seven errors made during translation into L2 were improper translations ['sgagnàr' was translated as 'masticare' (to chew) instead of 'mordere' (to bite)]; one error was a calque ['intivàr' was translated as 'intivare' (non-existent word in standard Italian) instead of 'azzeccare' (to guess)] and one error was a failure to respond.

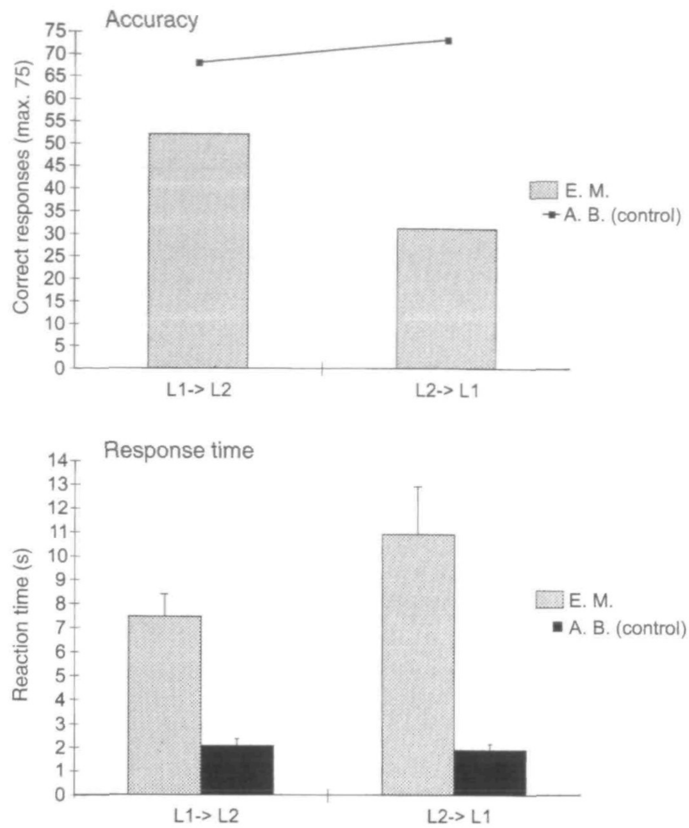

Fig. 2 Word translation tasks. Accuracy and response times are shown in the upper part and lower part of the figure, respectively; an analysis of variance (ANOVA) on response times with completely randomized design gave $F(3,220) = 29.33$, $P < 0.001$; the effect is due to the fact that EM was significantly slower during translation from L2 into L1 than from L1 into L2. There were no significant differences in the control subject's response time, comparing the two directions of translation. However, the control subject achieved a slightly higher accuracy when translating into L1. Columns represent means. The SE is indicated above each bar.

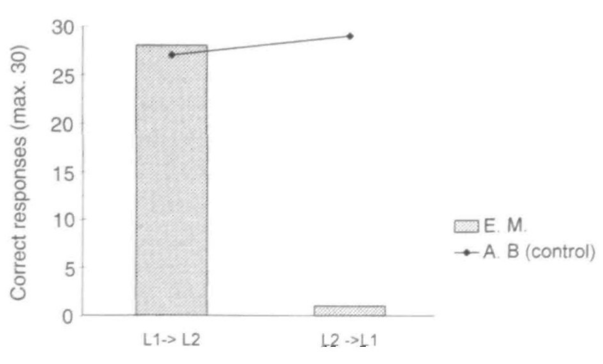

Fig. 3 Sentence translation tasks; accuracy of response of Patient E.M. and control subject A.B.

Sentence translation tasks

Figure 3 shows the accuracy of E.M. and A.B. (control) in the sentence translation task. Qualitative analysis of E.M.'s performance in this task is as follows: In the translation of sentences from L2 into L1, the patient repeated the sentences or paraphrased them in L2 [e.g. sentence 6 from Appendix 2, 'egli ha il vizietto che gli piace bere' (he likes drinking) was translated as 'avere il vizietto che gli piace bere' (to like drinking)]. When E.M. was asked to translate from L1 into L2, only two sentences were incorrect. In one sentence (sentence 26 in Appendix 2) there was one omission of free-standing grammatical morphemes ['la méta sta matita in scarsèla' (put this pencil in your pocket) was translated as 'mettere matita in tasca' (to put a pencil in one's pocket)]. The other was simply not translated. In this task, A.B. made one error which concerned only one word. In summary, an asymmetrical translation ability was present in E.M. [$\chi^2(1) = 45.17$, $P < 0.0001$] but not in the control subject. Furthermore, E.M. turned out to be impaired with respect to A.B. in translation from L2 into L1 [$\chi^2(1) = 48.6$, $P < 0.0001$] but not in the opposite direction ($\chi^2 < 1$).

Writing tasks

Theoretically, languages relying on both oral and written codes can be much more resistant to insults than languages based only on oral codes. Although, as previously mentioned, there are several examples of books and poems written in Venetan, this language is mainly oral. In addition, several differences between L1 and L2 are related to the different intonation features of the two languages. We reasoned that E.M.'s asymmetry in the direction of translation (and also her general linguistic impairments), if based on the number of available codes, should be different in oral and written tasks. To test this hypothesis we asked E.M. to do a written translation from L1 into L2 (and vice versa) of the same list of 75 words she had previously translated orally. The order of the words was randomized with respect to the oral translation sessions. When asked to translate into L1, E.M. scored 26 correct responses (versus 31 in the analogous oral task, $\chi^2 < 1$); 20 words were simply copied from the source language and 22 words were not translated; E.M. also made one calque, four literal paragraphias, one neologism and one circumlocution in L2. When asked to translate into L2, E.M. scored 54 correct responses (versus 52 in the analogous oral task, $\chi^2 < 1$); she made two literal paragraphias in L2, two semantic paragraphias [e.g. she translated 'ronchesàr' (to snore) as 'rombare' (to roar) instead of 'russare'], two circumlocutions in L2, three repetitions of the source stimulus and she failed to respond 12 times. In summary, the asymmetry in translation direction and the kind of errors involved were similar in written and oral tasks.

Follow-up study

Since changes over time in the pattern of linguistic deficits are greater in bilingual than in monolingual aphasics (Paradis, 1989), the follow-up study in our patient was theoretically fairly important. This study was conducted ~5 years after E.M.'s stroke. Testing took place at E.M.'s house in the presence of her husband and a young lady who looks after them. From a preliminary interview it was established that E.M. continued having troubles in speaking Venetan and preferred using standard Italian, even though all the people in her environment spoke in Venetan. No signs of mental deterioration were apparent and none were reported by E.M.'s relatives. The patient was given a standard spontaneous speech task ('describe how you make pasta with sauce') in both L2 and L1. The spontaneous speech task was given first in L2. A 10-min warm-up conversation in L1 followed, between E.M. and the examiner. Then, the patient was asked to perform the spontaneous speech task in L1. The analysis of spontaneous speech is shown in the right part of Table 2. As seen by the percentage of words in the non-requested language, E.M.'s tendency to speak in L2 in this follow-up session was similar to that observed 1 year after the stroke. Thus, the pattern of results had not changed drastically over 5 years. However, in the follow-up session, there was a tendency towards improvement in L2 which was not paralleled by a similar improvement in L1. For example, while the percentage of perseverations in L1 was higher in the follow up than in the sessions performed 1 year after the stroke, the opposite was the case for perseverations in L2. Moreover, neologisms, paraphasias and paragrammatisms were very rare, or even absent, 5 years after the stroke. Another difference between the two testing epochs concerned E.M.'s reduction of verbal fluency which was much stronger for L1 than for L2. Indeed, when speaking in L1, E.M. uttered 72.7 words per minute 1 year after the stroke and 52.3 words per minute 5 years after the stroke; in contrast, such values were 78.4 and 70.7 for L2.

Translation tasks were also given to the patient. In particular, she was asked to translate a list of 35 words from L1 into L2; these words were chosen from the same list used 1 year after the stroke. Finally, after a 10-min rest period, E.M. was asked to translate 28 of the above mentioned 35 words into L1. When E.M. reported not being able to translate a word, to avoid frustration, we gave her contextual prompts (e.g. a facilitation for the word wrinkle was 'things that appear on the face in elderly'); when such prompts were not effective, phonemic cues were also given (e.g. the examiner uttered the word's first phoneme). An error was scored when neither contextual nor phonemic cues induced correct responses. Response times for each correct response were recorded by means of a manually controlled stop-watch. The order of the translation tasks was scheduled to minimize E.M.'s dissociation. Note, in fact, that words in L1 presented to E.M. by the examiner in the L1 to L2 task may have primed the subsequent translation in the opposite direction. Thus, once again an asymmetrical translation pattern in favour of L2 is to be considered a very robust result. While E.M. scored 23 correct responses (65.7%) out of 35 stimuli in translation of words into L2, she scored only 11 correct responses (39.3%) out of 28 stimuli in the opposite direction [$\chi^2(1) = 4.37$, $P = 0.03$]. The number of errors was also significantly higher in the L2 to L1 than in the L1 to L2 translation [one out of 35 (4.3 %) versus eight out of 28 (28.6 %), $\chi^2(1) = 8.39$, $P < 0.003$]. In the L1 to L2 translation, eight contextual and four phonemic cues brought about correct responses. In the L2 to L1 translation, three contextual and four phonemic cues brought about correct responses. It is worth noting that, on several occasions, E.M. was not even able to translate words into L1 when the examiner had uttered the desired word only 25 min before. Such an asymmetrical performance cannot be due to a speed-accuracy trade-off, sincer E.M. was not only more accurate when translating into L2 but she was also more rapid in that direction of translation. Indeed, her average response time for correct responses was 7.5 s (±1.7 SD) in the L1 to L2 translation and 11.7 s (±3.5 SD) in the L2 to L1 translation.

Discussion

In spite of an increasing number of reports on subcortical aphasia in monolinguals, cases of bilingual subcortical aphasia are still rare in the literature (Paradis, 1995). Patient E.M. is the first case of bilingual subcortical aphasia in whom an extensive neurolinguistic analysis has been carried out. Her deficit consists of a paradoxical recovery of L2, a language that the patient had been using only occasionally in her life. It is also remarkable that such a pattern of recovery was rather stable from 1 to 5 years after the stroke. The patient's aphasia was qualitatively similar in both L1 and L2. This result does suggest that the notion of totally separate neural bases for different languages is rather a theoretical one.

Patient E.M.'s recovery differs from standard patterns of recovery where the different languages are recovered in parallel, or where the mother tongue (so-called Ribot's rule), or the most familiar language (so-called Pitre's rule) are preferentially recovered (Paradis, 1977, 1989; Albert and Obler, 1978). The possible role of affective factors in the selective recovery of L2 (Minkowski, 1927, 1964) is excluded by the fact that E.M. did not have any particular affective relationships with people speaking standard Italian. The influence of the linguistic environment soon after the stroke (Bycohwsky, 1919) cannot explain E.M.'s recovery either, because patients and staff at the hospital where the patient was admitted typically spoke in Venetian. The hypothesis that writing and reading processes may bias the recovery of one language instead of another (Halpern, 1941) deserves some comment. Standard Italian is based on both written and oral codes. By contrast, Venetan is mainly an oral language, even though there are texts (especially poems) written in Venetan. However, E.M. was not used to reading texts in Venetan. It is thus possible that E.M.'s selective recovery of L2 is related to the possibility of using both oral and written codes. Two lines of evidence, however, do suggest that such interpretation may not be adequate. The first is that E.M.'s experience of reading and writing standard Italian is rather low. The second, and possibly more important, is that E.M.'s asymmetrical translation pattern (worse from L2 into L1 than vice versa) was fairly similar in both written and oral tasks.

Some patterns of recovery in bilinguals may be best explained by referring to the different strategies used in learning a given language. Gelb (1937) described a patient who used a language learned in formal sets by conscious application of formal rules in order to recover a language mastered in natural learning sets without any conscious strategy. This patient, a professor of Latin, was unable to speak in his mother tongue (German) as a consequence of a lesion presumably centred on the left frontal lobe. By contrast, the patient was able to speak Latin, a language he had studied formally as an adult. The patient reported that he was able to significantly recover his mother tongue by translating words and sentences from the Latin into German. So, although anecdotal, this report suggests that impairment of the most automatized language may force the use of a less practiced second language. Paradis (1994) recently proposed that bilinguals may master each language by using different sets of implicit and explicit memory systems which, as already mentioned, rely upon largely separate neural structures (Saint Cyr *et al.*, 1988; Perani *et al.*, 1993; Ostergaard and Jernigan, 1993; Shimamura, 1993; Butters *et al.*, 1994; Salmon and Butters, 1995; Schacter, 1995; Squire and Knowlton, 1995). Mother tongues, typically acquired early in life, are highly automatized and mainly used through unconscious procedures. In contrast, the other languages, which are generally acquired later in life, are typically learned and used through the conscious application of grammatical rules. Paradis (1994) maintained that while mother tongues may mostly rely upon implicit memory systems, the other languages mainly use explicit memory systems.

Therefore, a possible explanation for the unusual deficit of E.M. may be the following: a lesion centred on the left basal ganglia, which are part of implicit memory systems, mainly affects the most automatized language; in contrast, E.M.'s less practiced language, possibly relying on other cortical and subcortical structures, is selectively spared. A similar explanation may apply to the patient described by Gelb (1937) especially considering the tight relation between left basal ganglia and left frontal cortex.

Evidence for a differential cerebral representation of different linguistic information has been provided using the technique of event related potentials. Neville *et al.* (1992) examined event-related potentials elicited by words providing semantic information (open class) or grammatical information (closed class) in English monolinguals and deaf people whose mother tongue was a gestural sign language and who had learned standard English as a second language later in life. In monolinguals, while event-related potentials indices of semantic processing were distributed over the left temporo-parietal lobes, indices of grammatical processing were distributed over the left frontal lobe. A different pattern of cortical potentials was evoked in deaf people where both closed and open class words selectively activated the left temporo-parietal lobes. Neville *et al.* (1992) concluded that acquisition and automatization of grammatical rules require early exposure; these authors also suggested that systems for semantic and grammatical processes have a different maturational time course. Both electrophysiology (Neville *et al.*, 1992) and clinical data suggest that closed class words, which are highly automatized, are mostly represented in the left frontal lobe (Friederici and Saddy, 1993), a neural structure directly connected with the basal ganglia; the frontal lobe–basal ganglia connection may be a relevant part within the complex subcortical–cortical network involved in automatizing motor and cognitive abilities (Alexander *et al.*, 1986; Wallesch and Papagno, 1988; Graybiel, 1995; Salmon and Butters, 1995). Moreover, there is some recent evidence to suggest that the basal ganglia may be involved in automatic speech production. Speedie *et al.* (1993) reported on a bilingual patient (mother tongue, French; second language, Hebrew), who suffered from a subcortical haemorrhage involving the right basal ganglia. Propositional speech was unaffected. However, the patient was impaired in producing, but not in understanding, automatic speech; indeed, he was no longer able to sing, swear, use conventional social greetings and conversational fillers, or recite overfamiliar verses and rhymes. The linguistic deficit, similar in both French and Hebrew, paralleled an impairment in the Tower of Hanoi and the stylus maze-tracing tests, thus supporting the notion of a close relationship between automatic language, the basal ganglia and implicit memory.

Patient E.M.'s deficit may also be considered as a pathological fixation on a foreign language from a defective putative mechanism for switching across languages. According to Poetzl (1925, 1930), such a mechanism is located in the left supramarginal gyrus of bilingual and polyglot subjects. Apart from the fact that switching mechanisms may not be different for linguistic and non-linguistic behaviour, and therefore not be specific to the bilingual brain (Paradis, 1989), studies have suggested that the neural bases of choice behaviour are most likely to reside in the prefrontal and frontal cortices (Zatorre, 1989; Gray *et al.*, 1991). However, given the close relationship between these areas and the basal ganglia (Alexander *et al.*, 1986; Wallesch and Papagno, 1988; Zatorre, 1989; Gray *et al.*, 1991), E.M.'s disruption may fit with the suggestion that the basal ganglia are an important node in the network for changing from one behaviour to another (Graybiel, 1995). In addition, it is noteworthy that E.M.'s performance in the Wisconsin card sorting test, a non-verbal task which taps the ability to change from one criterion of choice to another, was within normal range. This result suggests that E.M.'s fixation behaviour is mostly linguistic in nature.

Impairments in cross-language translation, which are peculiar features of bilingual and polyglot aphasia, may selectively affect only one direction (e.g. only from L1 into L2 or vice versa) or both directions of translation (Paradis, 1984). Also, bilingual aphasics may present with a compulsory tendency to translate their spontaneous speech (Kauders, 1929; Perecman, 1984; Lebrun, 1991). Translation without comprehension, i.e. correct translation of a verbal command which was not understood, has also been reported

(Veyrac, 1931). The phenomenon of paradoxical translation, i.e. the ability to translate into a language which is unavailable for other tasks, like comprehension or spontaneous speech, is an intriguing phenomenon (*see* Paradis *et al.*, 1982). Such a phenomenon strongly supports the hypothesis that translation processes may be quite distinct from those accounting for other linguistic skills, and that these processes may also be present within each system. It could be the case, for example, that separate functional modules exist for each direction of translation (Paradis, 1984). Unlike those in the patients described by Paradis *et al.* (1982), E.M.'s deficit was stable over years; in addition, her lesion was subcortical. However, E.M.'s translation pattern is somewhat paradoxical. Indeed, on both oral and written tasks, she was able to translate into L2 with a significantly higher proficiency than into L1. Studies in people without brain damage have shown that translation into one's mother tongue is usually easier than into a second language (Fabbro *et al.*, 1990, 1991); the performance of E.M.'s husband, who tended to be better at translating into his mother tongue, is in keeping with these studies. Echolalic responses seem to ensue from the speakers' inability to inhibit the motor interiorization of verbal expressions produced by their interlocutor. About one-third of echolalic utterances made by E.M. during spontaneous speech in L1 were spontaneously translated into L2. For example, when the examiner asked E.M. 'No ièra piu' bona de parlàr?' (were you no longer able to speak ?) the patient uttered the sentence in correct standard Italian ('Non era piu' capace di parlare?') instead of answering the question. Like spontaneous translation and translation without comprehension described in bilingual and polyglot aphasia (Veyrac, 1931; Perecman, 1984; Paradis, 1989), echolalic translation suggests a partial separation of translation and verbal comprehension processes (Paradis, 1995; Fabbro, 1996). However, E.M.'s performance does not necessarily hint at a deficit in select translation components; it may simply reflect the patient's impairment in mother tongue production.

Monolingual aphasics who suffered from lesions involving the left basal ganglia present not only with signs of non-fluent aphasia but also with verbal and semantic paraphasias and even comprehension deficits which are typically frequent in fluent aphasia (Damasio *et al.*, 1982; Wallesch *et al.*, 1983; Wallesch and Papagno, 1988, Démonet *et al.*, 1991; Crosson, 1992). Such a pattern of linguistic impairment was also evident in E.M., who (particularly 1 year after the lesion) showed not only grammatical errors but also some comprehension deficit in the token test as well as verbal and semantic paraphasias. The question arises: why does subcortical aphasia have typical symptoms of both fluent and non-fluent aphasia? Even though there is a lot of evidence to suggest the importance of subcortical structures in complex motor and cognitive behavior (Alexander *et al.*, 1986; Graybiel, 1995), several authors (Alexander, 1989; Bhatia and Marsden, 1994, Corbett *et al.*, 1994; Mega and Alexander, 1994; D'Esposito and Alexander, 1995) have reported that lesions in the basal ganglia affect cognitive functions mainly if there are concomitant lesions in the paraventricular white matter. A lesion to this last structure is likely to influence both anterior and posterior language systems by interrupting long-range, cortico-cortical and cortico-subcortical connections. This could explain the multifarious linguistic impairment following subcortical lesions. The notion of subcortical aphasia does not imply that subcortical structures work in isolation. Indeed, regional cerebral blood flow studies document both cortical (Perani *et al.*, 1987, 1988; Weiller *et al.*, 1993) and cerebellar (Perani *et al.* , 1988) areas of hypoperfusion in patients with subcortical stroke. These remote effects, prominent in the acute phase, support the notion that cognitive functions rely upon large cortical and subcortical neural networks. Although remote effects have not been investigated by means of functional imaging in E.M.'s case, such effects may have contributed to her syndrome. However, the cross-linguistic dissociation in E.M. was present a long time after the stroke, thus suggesting a relation between her linguistic behaviour and the subcortical lesion.

Acknowledgements

The patience and cooperation of E.M. and her husband are gratefully acknowledged. Thanks are due to Dr F. Ursino and Dr G. Marcato for their expert advice on the Venetan dialects, and to Dr V. Daro' for her comment on the manuscript. The financial contribution of the Consiglio Nazionale delle Ricerche and the M.U.R.S.T., Italy is also acknowledged.

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Received March 5, 1996. Revised April 11, 1996.
Accepted May 8, 1996

Appendix 1

(1) pisello = bìso (pea);
(2) spegnere = stuàr (to turn off);
(3) coperta = cuèrta (blanket);
(4) orecchio = récia (ear);
(5) prurito = spìra (itch)
(6) rabbrividire = sgrisolàr (to shiver)
(7) mangiatoia = grépia (manger)
(8) almeno = almànco (at least)
(9) bambino = butìn (child)
(10) prendere = tôr (to take)
(11) brivido = sgrisolòn (shiver)
(12) trovare = catàr (to find)
(13) ascelle = leséne (armpit)
(14) falegname = marangòn (carpenter)
(15) colino = passìn (strainer)
(16) ingordigia = ingordìsia (greediness)
(17) ragazza = bùtela (girl)
(18) letame = luàme (manure)
(19) uccidere = copàr (to kill)
(20) scrofa = ròia (sow)
(21) lenzuolo = nisòl (sheet)
(22) ubriacarsi = inciucàr (to get drunk)
(23) tasca = scarsèla (pocket)
(24) coltello = cortèl (knife)
(25) oggi = ancò (today)
(26) accorciare = scurtàr (to shorten)
(27) suocero = missèr (father-in-law)
(28) spossatezza = lèla (weariness)
(29) azzeccare = intivàr (to guess)
(30) sedia = caréga (chair)
(31) tacchino = dìndio (turkey)
(32) bicchiere di vino = gòto (glass of wine)
(33) strappare = sbregàr (to snatch)
(34) bambola = pùa (doll)
(35) ruga = ràpola (wrinkle)
(35) stirare = sopressàr (to iron)
(37) fetta = slèpa (slice)
(38) gelso = moràr (mulberry-tree)
(39) dietro = indrìo (behind)
(40) coprire = quaciàr (to cover)
(41) febbre = féara (fever)
(42) spazzatura = sgaùia (garbage)
(43) bestemmiare = ostiàr (to curse)
(44) fretta = prèssia (rush)
(45) moglie = moiér (wife)
(46) sinistra = sànca (left)
(47) spruzzare = sbrofàr (to sprinkle)
(48) pepe = péar (pepper)
(49) lepre = léoro (hare)
(50) troppo = màssa (too much)
(51) sabato = sàbo (Saturday)
(52) succhiare = ciuciàr (to suck)
(53) uccello = osél (bird)
(54) spinta = sburtàda (push)
(55) tossire = sbossegàr (to cough)
(56) vicino = arènte (nearby)
(57) pantaloni = bràghe (trousers)
(58) siepe = sésa (hedge)
(59) russare = ronchesàr (to snore)
(60) scappellotto = scufiòto (slap)
(61) scolapiatti = sgossaròla (draining-board)
(62) soldi = schèi (money)
(63) picchiare = ciocàr (to hit)
(64) promesso sposo = noìsso (fiancée)
(65) guancia = ganàssa (cheek)
(66) gonna = còtola (skirt)
(67) mordere = sgagnàr (to bite)
(68) noce = nosàr (walnut-tree)
(69) salvadanaio = musìna (moneybox)
(70) bottiglia = bòssa (bottle)
(71) somaro = mùsso (donkey)
(72) affrettarsi = spessegàr (to hurry up)
(73) forchetta = piròn (fork)
(74) merda di mucca = boàssa (cow shit)
(75) tazzina = chìchara (small cup)

Appendix 2

(1) Ho bisogno di vederti e di parlarti = Mì bisòn che te veda e che te parla
(I need to see and talk to you)
(2) Guardare una ragazza = Ociàr 'na bùtela
(To look at a girl)
(3) Fare un sonnellino = Fár la pìsa
(To take a nap)
(4) I bersaglieri hanno il cappello col fiocco = I bersaliéri i g'à la baréta col fiòcolo
(The bersaglieri have a flock on their hat)
(5) Le ha dato un pizzicotto sulla guancia = La ghe dà 'n pissegón su la ganàssa
(He pinched her on her cheek)
(6) Ha il vizietto che gli piace bere = El g'à el vissiéto che ghe piàse el gòto
(He likes drinking)
(7) L'hanno licenziato perché rubava = L'à mandà via parché l'à gratà
(He was fired because he was used to steal)

(8) Signore, aiutami, non ne posso più! = Signor, iutème, no gh'in posso più!
(God, help me, I'm exhausted)
(9) Il vino è il latte dei vecchi = El vin l'è el làte dei vèci
(Wine is milk for the elderly people)
(10) I panni sporchi si lavano in casa = Le ròbe spórche se le lava in casa
(Don't wash your dirt linen in public)
(11) Ella ha una lingua che sembra una spada = La g'à 'na léngua che pàr 'na spàda
(She has the tongue like a sword)
(12) Piange ed è bianca come un lenzuolo = La piànse e l'è biànca come un nisòl
(She is crying and is pale like a linen)
(13) Quel che è troppo è troppo = Quel che è màssa è màssa
(Enough is enough)
(14) Il merlo canta dove ha il nido = El mèrlo el canta dove el g'à el nio
(The blackbird sings where his nest is)
(15) Hai la gonna che pende da un lato = Te gh'e la còtola che pincòna da 'na pàrte
(Your shirt is showing on one side)
(16) La ragazza mi è sembrata buona = La putéla la mà pàrso bòna
(The girl seemed to be good)
(17) Avere un sassolino nella scarpa = Averghe 'n sasséto in te 'na scàrpa
(To have a stone in one's shoe)
(18) Hai piacere che finisca in fretta? = Gh'èto piasér che la fenìssa en préssia?
(Do you like him/her to finish soon?)
(19) Da ragazza ho avuto il fidanzato = Mì gò vù 'l mòroso da butéla
(As a girl I used to be engaged)
(20) Hai finito? = Eto finìo?
(Are you done?)
(21) Sembra che il tempo si rimetta = Pàr che 'l tempo el se rimeta
(It looks like the weather is settling)
(22) Si è fatto coraggio e ha corso il rischio = El s'à fàto coràio e l'l'à risciàda
(He plucked up courage and ran the risk)
(23) Per lei perderei la testa = Mì par éla perdarìa el çervèl
(I would loose my head for her)
(24) Aveva un febbrone che vaneggiava = El g'avea 'na féara che 'l savariàva
(He was raving in fever)
(25) Egli spinse il portone che era là vicino = El sbùrtá el portón che gh'era là arènte
(He pushed the door that was next to him)
(26) Metta questa matita in tasca! = La méta sta matita in scarsèla
(Put this pencil in your pocket)
(27) Si metta seduto! = La se méta in sentòn!
(Sit down!)
(28) Ha sgridato la cassiera = El ga dà 'na sigàda a la cassiéra
(He reproached the cashier)
(29) Basta che si guardi allo specchio = Basta che se la vàrda éla in t'el spèio
(Suffice for him to look at himself in the mirror)
(30) Presto ti metterò L'anello al dito = Presto la véra mì te méto in déo
(I'll soon put a ring on your finger)