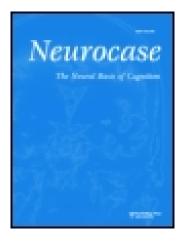
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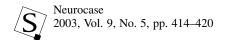
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Dissociation Between Personal and Extrapersonal Neglect in a Crossed Aphasia Study

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Abstract

Several reports document crossed aphasia following a right cerebral infarct. However, few of them provide a detailed investigation of associated neuropsychological disorders. A personal neglect disorder with no difficulty in orienting attention in the contralesional space has not been frequently reported independently of lesion side for the language deficit. In most cases, the deficit is described in the acute period.

We report the case of a patient who showed severe crossed aphasia several months after cerebral damage. In addition to his language deficit, he suffered from finger agnosia, acalculia, and right/left confusion. Although he was able to orient attention in the contralesional space, he had a persistent personal neglect disorder with severe difficulty in attending to his own body. Results suggest that right hemisphere language dominance does not preclude ipsilateral specialisation for other functions. Furthermore, the dissociation between the two spatial functions provides further confirmation that they are subserved by two independent systems.

Introduction

In right-handed people, a language deficit due to a right hemispheric lesion is rare (Albert et al., 1981). However, there are several well-documented cases of aphasia following right cerebral damage (crossed aphasia) in the literature (Trojanowski et al., 1980; Hindson et al., 1984; Kapur and Dunkley, 1984; Basso et al., 1985; Alexander et al., 1989; Faglia and Vignolo, 1990; Rey et al., 1994; Stefanis et al., 1997; Nagaratnam et al., 1999). Often the language deficit recovers quickly in time (Ludwig, 1939; Urbain et al., 1978; Henderson, 1983; Basso et al., 1985; Roebroeck et al., 1999). However, cases of a persistent language deficit have also been reported (Trojanowski et al., 1980; Hindson et al., 1984; Kapur and Dunkley, 1984; Basso et al., 1985; Walker-Batson, 1988; Faglia and Vignolo, 1990; Rey et al., 1994). In some cases, the language disorder was associated with visuospatial deficits, suggesting that, also in subjects with inverted hemispheric dominance, the right hemisphere is responsible for both verbal and visuospatial functions (Henderson, 1983; Kapur and Dunkley, 1984; Basso et al., 1985; Faglia and Vignolo, 1990). However, few crossed aphasia studies have provided a detailed investigation of the variety of neurobehavioural disorders that can occur after right hemisphere damage (Aboo-Baker and Labauge, 1987; Castro-Caldas et al., 1987; Fournet et al., 1987; Nagaratnam et al., 1999).

Regardless of the side of the hemispheric localisation for language, the most frequent deficit described after right

cerebral damage is unilateral neglect (Hècaen and Angelergues, 1961; Gainotti et al., 1972). Patients with this syndrome are unable to detect and orient attention to stimuli in the contralesional space (extrapersonal neglect) and/or toward the contralesional hemisoma (personal neglect). Patients with personal neglect but no impairment for extrapersonal space are quite rare (Bisiach et al., 1986; Pizzamiglio et al., 1989a; Guariglia and Antonucci, 1992; Peru and Pinna, 1997). As far as we know, no cases of crossed aphasia and personal neglect have been reported. Some authors attribute the deficit to an attentional disorder that causes inability to attend to the side of the body contralateral to the lesion (Kinsbourne, 1995; Semenza, 2001). Others refer to autotopoagnosia (AT) as a possible related sign of the syndrome (Coslett, 1989, 1998; Guariglia and Antonucci, 1992). It has been argued that AT is a function in the domain of the body structural description system (Sirigu et al., 1991; Buxbaum and Coslett, 2001). Patients with AT suffer from deficits in the representation of local relationships between body parts. Recently, Denes and colleagues described two AT patients with language disorders. They hypothesised that AT may be due to a lesion in a neural circuit located in the language dominant hemisphere (Denes et al., 2000). However, in most of the cases cited personal neglect recovered quite rapidly and was present only in the acute phase (Peru and Pinna, 1997). As far as we know, only one study

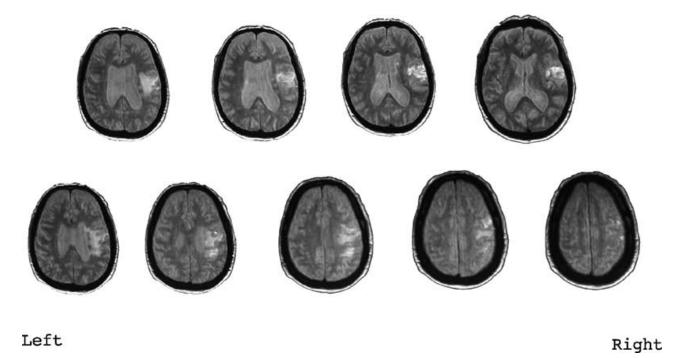


Fig. 1. GR's MRI scan evidences a right frontal-parietal and temporal insular cortical and sub-cortical lesion.

(Guariglia and Antonucci, 1992) reported the case of a patient with severe personal neglect that persisted many months after the onset of cerebral damage.

In this paper, we present the case of a right-handed patient who developed crossed aphasia and left hemiplegia after an infarct of the right hemisphere. To better understand the patient's right hemisphere competence for different cognitive functions, he was submitted to a thorough neuropsychological investigation. Although language deficits were present, they did not prevent us from showing a clear dissociation between a severe personal neglect and lack of signs of alteration in his ability to orient attention to the contralesional space. Furthermore, contrary to most previous findings, both the language and the personal neglect disorder were present many months after onset of the right cerebral lesion.

Case report

GR, a 78-year-old, right-handed man with five years of education, was referred to our hospital on February 22, 2001 for motor and language rehabilitation. On October 30, 2000, he had a cerebral ischaemia that left him with left-sided hemiparesis and aphasia. A neurological examination was administered using a standard procedure with a four-point scale ranging from 0 (no deficit) to 3 (severe deficit). Motor impairments were very slight (score of 1) for both the upper and the lower left limb. However, the patient complained about these deficits, showing that he was aware of his illness.

There were no signs of hemifield or hemisensory defects. His language production was severely reduced, with mildly impaired comprehension of verbal material. An MRI scan performed four months after the event showed the presence of right frontal-parietal and temporal insular cortical and sub-cortical lesions, confirming that the infarct was localised exclusively in the right hemisphere (see Fig. 1).

Handedness was evaluated by means of the Edinburgh Inventory Test (Oldfield, 1971); the patient's laterality score was 12/12 in favour of the right hand. He had no family history of left-handedness.

Neuropsychological investigation

GR was examined four months post-onset. At the time of the experimental investigation (March, 2001), he underwent a series of neuropsychological tests. Language was assessed using the Esame del Linguaggio II (Ciurli et al., 1996). Spontaneous speech was totally absent. A severe verbal apraxia prevented him from responding on all production tasks, such as oral naming, repetition and reading. His writing ability was also severely affected. Both writing words on dictation and writing on a picture naming task were characterised by sequences of graphemes (non words), which showed no relationship to the target stimulus. Automated sequences (counting from 1 to 10, naming the days of the week and months of the year) were impaired. His verbal comprehension was very well preserved both for single words and commands (Table 1). However, he scored 14 on the Token test (cut-off score 29/36; De Renzi and Faglioni, 1978).

Due to the presence of verbal apraxia, his verbal memory span was not tested. His spatial memory span was low on the Corsi test (span: 4) but in the normal range for his age and educational level (Orsini *et al.*, 1987). He showed no oral,

Table 1. GR's correct responses on the language test (Ciurli et al., 1996)

	Picture description	Noun naming	Verb naming
Production tasks			
Oral	_	0/20	0/20
Written	_	0/20	0/20
	Unrelated words	Semantic rel. words	Commands
Comprehension tasks			
Oral	20/20	19/20	20/20
Written	20/20	19/20	19/20
	Words	Non-words	Sentences
Repetition	0/20	0/20	0/20
Reading	3/20	0/20	0/20
Writing on dictation	0/20	0/20	0/20

Legend. — the patient did not produce any words. Semantic rel. words = 5 words belonging to the same semantic category (total of 4 categories).

ideomotor, or ideational apraxia (De Renzi *et al.*, 1966, 1968, 1980). Scores on Raven's Coloured Progressive Matrices test and on the Constructive apraxia test were at the lower end of the normal limits (18/36, cut-off 18; Basso *et al.*, 1987 and 8/14, cut-off 8, Spinnler and Tognoni, 1987, respectively) but in the normal range for his age and educational level.

To sum up, the language test revealed the presence of severe non-fluent aphasia with verbal apraxia and relatively good ability to understand verbal material. Although the patient underwent language rehabilitation five times a week, a reassessment of his language function two months later (May, 2001) showed no significant improvement on any tasks.

The patient underwent another series of tests. Although he was able to perform a number comparison task, he had great difficulty in making very simple calculations (EC301 Calculation Battery, Deloche and Seron, 1991). To test his ability to discriminate between left and right, we presented him with a street maze on a sheet of paper with different crossing points marked by red dots. The patient was asked to follow the path with a pencil and to stop when he reached the red dot. Then, the examiner asked him to turn either toward the right or the left side of the maze. GR made a total of 33 errors in 50 trials, demonstrating severe right/left confusion.

GR was asked to perform a Finger Agnosia test (Guariglia et al., 2002). The examiner touched one finger at a time on each hand and the patient was asked to indicate the finger stimulated on a hand drawn on a sheet of paper. The stimulation was made in three different conditions: (1) with hands covered, palms down and fingers fully extended and spread, (2) with hands covered, palms up and fingers fully extended and spread, (3) with hands closed and covered and fingers and thumb partially flexed and adducted so that they almost touched. In all conditions, GR's performance was clearly defective for both the right and the left hand. Results in all conditions are reported in Table 2.

The above results suggest that the patient was suffering from acalculia, right/left confusion and finger agnosia

Table 2. GR's correct responses on the Finger Agnosia test

Pointing	Correct responses	
Extended hand palm down		
Right hand	8/15	
Left hand	8/15	
Extended hand palm up		
Right hand	8/15	
Left hand	4/15	
Closed hand		
Right hand	5/15	
Left hand	5/15	

Table 3. GR's correct responses on the Heminattention battery (Pizzamiglio *et al.*, 1989)

Tests	Number of correct responses
Letter cancellation	Left 37/53, Right 34/51
Albert test	Left 11/11, Right 10/10
Wundt/Jastrow (unattended responses)	Left 18/20, Right 20/20
Line bisection	3/3
Star cancellation	Left 27/27, Right 27/27
Personal neglect ^a	5/9

^aZoccolotti and Judica, 1991.

which, together with his writing disorders, could indicate Gerstmann's Syndrome.

The patient was then subjected to a screening battery of tests for extrapersonal neglect (Pizzamiglio *et al.*, 1989b). He made no errors on the line cancellation test (Albert, 1973) or on the star cancellation test (Wilson *et al.*, 1987). On the letter cancellation test (Diller and Weinberg, 1977), he made 16 omissions on the left side of the paper and 17 on the right side, showing no signs of extrapersonal neglect. He correctly bisected three lines out of three. On the Wundt-Jastrow Illusion test (Massironi *et al.*, 1988), he gave two unattended responses when the stimulus was oriented toward the left and none when it was oriented toward the right. This performance was in the normal range (Table 3).

GR was then administered a Semi-structured Functional Evaluation Scale with a four-point scale ranging from 0 (normal performance) to 3 (severe deficit) (Zoccolotti and Judica, 1991), which showed the presence of personal neglect. In particular, when asked to put on eyeglasses, his score was 3 because he failed to put the left bar over his ear and to centre the left lens over his eye. He also failed to shave his left cheek, obtaining a score of 2 (Table 3).

To further explore the patient's ability to use objects on his own body, he was presented with seven more objects (a mask to protect his eyes from the light, hand cream, gloves, nail scissors, cotton, earphones, and braces). GR failed to put on the left glove, the left brace, and the left earphone. While cutting the nails on his left hand, he omitted two nails and spent much more time cleaning his left ear than his right.

Although these were not all standardised objects, the results can be taken as further confirmation of GR's severe personal neglect.

To sum up, the results of the neuropsychological investigation showed that GR had severe non-fluent crossed aphasia. Together with his language deficit, the patient also presented Gerstmann's Syndrome, with severe difficulty in resolving calculation problems, right/left confusion, finger agnosia, and writing disorders. Finally, there was a dissociation between the patient's preserved ability to orient attention to the contralesional space and his severe difficulty in correctly using objects on the left side of his body (personal neglect).

As stated in the introduction, in some studies personal neglect has been interpreted as the consequence of autotopoagnosia (Coslett, 1989, 1998; Guariglia and Antonucci, 1992). Other studies suggest that the encoding of body position is represented in the language dominant hemisphere (Denes *et al.*, 2000).

Therefore, as a last step we investigated whether the finger agnosia deficit reflects a partial loss of spatial knowledge of body parts.

Word-picture matching task

First of all, we tested the patient to see if he was able to understand names belonging to the body category. For this purpose, we used the body pictures of The Italian Semantic Battery test (Laiacona *et al.*, 1993). The patient was asked to perform a word-picture matching task. For each body part named orally by the examiner, he had to point to the correct picture out of five semantically related alternatives. The patient made no errors (10 out of 10 correct responses).

Test of autotopagnosia (Semenza and Goodglass, 1985)

We tested 18 body parts. GR had to point to the body parts in three different conditions. In the oral condition, the examiner said the name of a body part and asked the patient to touch the part on his own body, on a human picture and on a multiplechoice sheet. In the visual condition, the examiner showed one body part picture at a time and asked the patient to point to the same part on his own body and on a picture. In the tactile condition, the examiner touched different body parts of the patient, while the latter had his eyes closed; then the patient, with eyes open, had to point to the same part on the human picture and on a multiple choice sheet. Two other conditions were tested. In the first one, the examiner pointed to a body part on a human picture and the patient had to touch the same part on his own body. In the second condition, the examiner pointed to a body part on his body and the patient had to touch the same part on himself. Errors were grouped into three categories: conceptual (i.e. index finger substituted for ring finger), contiguity (errors in faulty reaching, such as neck substituted for chin) and random (i.e. knee substituted for ear).

GR made only 2% of errors in all conditions. Contiguity errors (1%) were more frequent than conceptual errors (0.60%) and there were no random errors (control subjects perform at ceiling on this task (Piccardi *et al.*, 2000). Although he was free to touch either the right or the left side of his body, the patient's pointing was equally distributed between the two sides.

He was also administered a modified version of Semenza and Goodglass's test (1995). Here, the patient was explicitly asked to point to different parts of the left and right side of his body. GR made 24% of errors, which were equally distributed between the right and the left side. He never substituted one body part for another; errors were only due to right/left confusion (i.e. instead of pointing to the left knee, he touched the right one).

To sum up, the above results demonstrate that the patient was quite good at locating body parts. However, when explicitly asked to point to the left or the right side of the body, he often failed and his errors were all substitutions of the right with the left side and vice versa. This performance pattern is consistent with his right/left confusion deficit.

Test of body schema (Daurat-Hmeljiak et al., 1978)

This test for evaluating body representation in children was previously used to study body schema impairments in braindamaged patients (Guariglia and Antonucci, 1992; Guariglia et al., 2002). Test items include single tiles, each representing a part of the human body or the head. Two different tasks are performed: naming-localisation of the tiles and construction of the frontal and profile view of the human body and head. Due to the patient's aphasia disorder, the naming condition was not recorded. In the localisation condition, the examiner asks the subject to put a tile on a test table on which the position of the head (for the body) or the contour of the face (for the head) is depicted. The examiner records the position of the body part chosen by the patient on an answer sheet by covering the test table with a transparent grid. Before presenting a new tile, the previous one is removed. In the construction condition, all tiles necessary to construct the frontal and profile view of the head and body are given to the subject. When the subject completes the task, the examiner reports each tile position on the answer sheet.

GR's performance was highly defective, corresponding to that of <6.5 year-old children both in frontal and profile views.

When asked to construct the human body in the frontal view, he omitted one hand and put the other one almost off the table (see Fig. 2a). The same performance was repeated when the patient was asked to draw a picture of the human body (see Fig. 2b). In the construction of the profile body view, he chose all frontal body parts (see Fig. 3), showing his inability to develop a profile picture. He then refused to continue the task and construct the lateral view of the head.

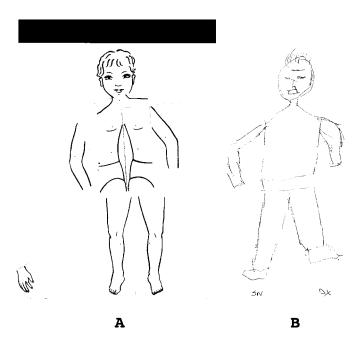


Fig. 2. Frontal (A) view reconstruction of the body. Drawing (B) of the human body.

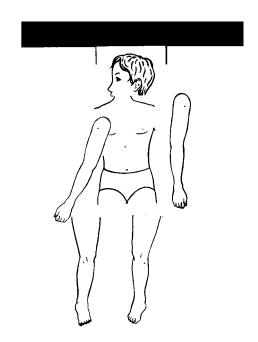


Fig. 3. Lateral view reconstruction of the body.

Discussion

In the present paper, we report the case of a right-handed patient, GR, who developed persistent neuropsychological deficits after a right cerebral infarct. He was affected by severe non-fluent crossed aphasia with finger agnosia, acalculia, and right/left confusion. In the spatial domain, he presented a dissociation between a severe personal neglect disorder and his preserved ability to orient attention to the contralesional space.

As stated in the introduction, although there are several documented cases of crossed aphasia in the literature (Trojanowski et al., 1980; Hindson et al., 1984; Kapur and Dunkley, 1984; Basso et al., 1985; Alexander et al., 1989; Faglia and Vignolo, 1990; Rey et al., 1994; Stefanis et al., 1997; Nagaratnam et al., 1999), few of them thoroughly investigate associated neurobehavioural disorders (Aboo-Baker and Labauge, 1987; Castro-Caldas et al., 1987; Fournet et al., 1987; Nagaratnam et al., 1999). Furthermore, in some cases the deficits described were present only in the acute phase (Ludwig, 1939; Urbain et al., 1978; Henderson, 1983; Basso et al., 1985; Roebroeck et al., 1999); thus, it is possible that the linguistic and spatial functions tested were only partially represented in the damaged right hemisphere. GR's neuropsychological impairments were present many months after disease onset (seven months), and although he underwent three months of language rehabilitation, the second language examination showed no signs of improvement on any tasks. It can be argued that the patient's lack of recovery in the spoken modality was partly due to his very severe verbal apraxia that prevented him from correctly performing the output tasks. However, there was also no recovery in the writing modality. Contrary to previous studies, his verbal apraxia was not associated with difficulty in performing bucco-facial movements. This raises the possibility that although there is a relationship between language and bucco-facial praxis (Alexander et al., 1989), their lateralisation is dissociable (Kertesz et al., 1984).

Another result was the patient's acalculia, right/left confusion and finger agnosia, which, together with his writing disorder, suggest that he may have developed Gerstmann's syndrome. This result is in line with previous studies which suggest that the syndrome is most often associated with lesions in the left dominant hemisphere (see Denes, 1999 for a review).

GR's ability to orient attention to the contralesional space was preserved. However, he had a severe personal neglect disorder that left him unable to use objects on the contralesional side of his body. To our knowledge, a dissociation between a preserved ability to orient attention to the contralesional space and a personal neglect disorder has never been reported in crossed aphasia studies. Furthermore, in contrast to most previous studies (Peru and Pinna, 1997), the deficit was persistent in time. Taken together, these results suggest that the two spatial functions may be represented by two independent systems. Guariglia and Antonucci (1992) reached similar conclusions in their single case study. We can also hypothesise that in our patient the two spatial functions are likely lateralised in two different hemispheres. Due to his very large infarct in the right hemisphere and to the lack of recovery of all functions tested, it can be argued that his ability to orient attention to the contralesional space was probably represented in the left intact hemisphere.

It has been suggested that personal neglect may be related to an autotopoagnosia deficit (Guariglia and Antonucci, 1992; Coslett, 1989, 1998) and that autotopoagnosia is represented in the language-dominant hemisphere (Denes *et al.*, 2000).

On Semenza and Goodglass's test (1985), GR was able to localise body parts, and on a modified version of the test (Semenza and Goodglass, 1985), his errors were all due to severe right/left confusion.

On the body schema test (Daurat-Hmeljak *et al.*, 1978), he had difficulty both in processing body spatial relations and in the construction of the body (see Figs. 2a and 3). Since the patient refused to perform other tasks and was discharged from hospital, we were not able to further assess his ability to perform body tasks. However, given his severe right/left confusion and his just above cut-off score on the constructional apraxia tests, his performance on the body schema task can likely be attributed to these concomitant symptoms.

In this context, GR's selective forgetfulness of both hands in the body schema construction as well as in the drawing (see Fig. 2a and 2b) should be observed. Given his finger agnosia deficit, it can be hypothesised that he suffered from a specific perturbation in the representation of this body part.

To sum up, two conclusions can be drawn from our results. First, they document a crossed aphasia study with a personal neglect disorder for the first time. Second, the presence of a dissociation between the patient's preserved ability to orient attention to the contralesional space and the personal neglect disorder further confirm that the two functions are subserved by different systems that may be represented in distinct cerebral hemispheres.

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Dissociation between personal and extrapersonal neglect in a crossed aphasia study

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Abstract

Several reports document crossed aphasia following a right cerebral infarct. However, few of them provide a detailed investigation of associated neuro-psychological disorders. A personal neglect disorder with no difficulty in orienting attention in the contralesional space has not been frequently reported independently of lesion side for the language deficit. In most cases, the deficit is described in the acute period.

We report the case of a patient who showed severe crossed aphasia several months after cerebral damage. In addition to his language deficit, he suffered from finger agnosia, acalculia and right/left confusion. Although he was able to orient attention in the contralesional space, he had a persistent personal neglect disorder with severe difficulty in attending to his own body. Results suggest that right hemisphere language dominance does not preclude ipsilateral specialisation for other functions. Furthermore, the dissociation between the two spatial functions provides further confirmation that they are subserved by two independent systems.

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Primary diagnosis of interest

Personal neglect disorder

Author's designation of case

GR

Key theoretical issue

 Functionally independent cerebral subsystems for orienting attention to the contralesional space and to the contralesional hemisoma

Key words: crossed aphasia; personal neglect

Scan, EEG and related measures

MR

Standardized assessment

Esame del Linguaggio II; Screening Battery of tests for Extrapersonal Neglect; Semi-structured Functional Evaluation Scale for Personal Neglect

Other assessment

Test of autotopagnosia; Test of body schema

Lesion location

• Right frontal-parietal and temporal insular cortical and sub-cortical lesions

Lesion type

Cerebral ischaemia

Language

Italian