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Dissociation between Position Sense and Visual-Spatial Components of Hemineglect through a Specific Rehabilitation Treatment*

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ABSTRACT

Current evidence suggests an association between contralesional extra-personal hemineglect and deficits of arm position sense in patients with damage to the right cerebral hemisphere. A unitary deficit may produce both disorders, or this association may reflect the anatomical contiguity of relevant brain structures. A rehabilitation treatment, devised for visual-spatial hemineglect, was used to investigate these hypotheses in 8 patients with damage to the right cerebral hemisphere. The treatment improved hemineglect, but not the position sense deficit. The severity of the latter was however transiently reduced by optokinetic stimulation, with effects similar to those found in visual-spatial hemineglect. These effects of rehabilitation suggest that extra-personal hemineglect and the neglect-related component of the position sense disorder of the left forearm are independent, though frequently associated, deficits. Implications for the design of rehabilitation programs are discussed.

A number of clinical and neuropsychological studies in patients with unilateral brain lesions suggest a close association between deficits of position sense, right brain damage, and visual-spatial hemineglect. First, most patients with damage to the right cerebral hemisphere and left visual neglect have deficits of position sense in the left arm, as assessed by a clinical neurological exam (Willanger, Danielsen, & Ankerhus, 1981). Second, according to a recent, community-based, epidemiological survey (Sterzi et al., 1993), the deficit of position sense is more frequent in patients with vascular lesions in the right hemisphere, as compared with patients with left hemisphere damage. Third, position sense disorders, assessed by a standardized experimental procedure, are much more severe in patients with lesions in the right hemisphere and left visual-spatial hemineglect, as compared with patients without hemineglect, independent of the side of the hemispheric lesion (Vallar, Antonucci, Guariglia, & Pizzamiglio, 1993a; Vallar, Guariglia, Magnotti, & Pizzamiglio, 1995).

In patients with left hemineglect, the impairment involves both the left forearm, contralateral to the side of the right hemisphere lesion, and the right ipsilateral forearm, although in the latter the deficit is less severe (Vallar et al., 1993a; Vallar et al., 1995). In patients without hemineglect, by contrast, the disorder is milder, and confined to the forearm contralateral to the side of the lesion, in line with the classic neurological views (Adams, Victor, & Ropper, 1997; Bickerstaff, 1973; Rowland, 1995). Finally, the deficit of position sense of patients with damage to the right hemisphere and left hemineglect is transiently improved by optokinetic stimulation.

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with a leftward (contralesional) direction of the movement, in both the left and the right forearm, whereas a stimulation with a rightward (ipsilesional) direction has negative effects (Vallar et al., 1993a; Vallar et al., 1995). This visual stimulation has similar direction-specific effects on left extra-personal visual-spatial hemineglect in patients with damage to the right cerebral hemisphere, asked to bisect horizontal lines (Pizzamiglio, Frasca, Guariglia, Incoccia, & Antonucci, 1990), or to point to the position they felt lay “straight ahead” of their bodies’ orientation (Karnath, 1996).

One interpretation (Vallar et al., 1993a; Vallar et al., 1995) of these findings is in terms of an ipsilesional distortion of a unitary spatial representation, which concerns both the position of body segments and objects in extra-personal space, and involves the left and the right half-spaces, in the horizontal and vertical dimensions. In patients with damage to the right hemisphere this distortion, in addition to producing the manifold manifestations of visual-spatial hemineglect, brings about a neglect-related defective perception of the spatial position of body parts, which, added to the primary sensory deficit, increases the severity of the position sense disorder. This view is based on a negative result, namely, the failure to find a dissociation between deficits of position sense, modulated by optokinetic stimulation, and visual-spatial hemineglect (Vallar et al., 1993a; Vallar et al., 1995).

It remains possible, however, that the representation of the spatial position of body segments (the body-in-space) differs from the egocentric representation of extra-personal space concerned with the spatial position of objects. This hypothesis should be taken into consideration because the neglect-related component of somatosensory deficits (defective perception of light touch) may occur in the absence of visual-spatial hemineglect and vice versa (Vallar, Bottini, Rusconi, & Sterzi, 1993b; Vallar, Rusconi, & Bernardini, 1996). No specific data concerning deficits of position sense are available, however. According to the latter view, the close association between visual-spatial hemineglect and disorders of position sense might be better explained in terms of close anatomical contiguity between relevant neural structures, which are likely to be conjointly damaged by a cerebral lesion.

In this study, we used rehabilitation of visual-spatial hemineglect as a tool to dissociate deficits of position sense from visual extra-personal neglect. Pizzamiglio et al. (1992), and Antonucci et al. (1995) set up a rehabilitation method which has significant positive effects on left extra-personal visual-spatial neglect. The improvement is selective, because the patients’ defective performance in a number of tests assessing visual-perceptual nonverbal abilities (face recognition, constructional apraxia, non-verbal reasoning) is not affected by the treatment.

If visual-spatial hemineglect and the neglect-related component of the deficit of position sense are produced by the distortion of a unitary system, concerning both body parts and objects in extra-personal space, both deficits should be improved by the rehabilitation treatment. By contrast, were discrete representational systems involved, the effect of rehabilitation should be confined to visual-spatial hemineglect.

METHOD

Subjects
Participants were 8 patients with damage to the right cerebral hemisphere and left visual-spatial hemineglect. All patients were right-handed and had no history of psychiatric disorders or dementia. In all patients the lesion was assessed by MRI. Table 1 summarizes the demographic, neurological, and neuropsychological features of the patients. Motor and somatosensory (upper and lower limbs), and visual half-field deficits (upper and lower quadrants) were assessed through a standardized neurological exam, with scores ranging from 0 (no deficit) to 6 (maximum deficit) (Bisiach & Faglioni, 1974). All patients were examined in a chronic stable phase, at least 60 days after stroke onset (Antonucci et al., 1995; Pizzamiglio et al., 1992).

Rehabilitation Treatment
The training program (Antonucci et al., 1995; Pizzamiglio et al., 1992) comprised 40 sessions (5 per week, for 8 consecutive weeks), each session
Table 1. Demographic and Neurological Features of Eight Patients with Damage to the Right Hemisphere and Left Visual-Spatial Hemineglect.

<table>
<thead>
<tr>
<th>Case</th>
<th>Age/Sex</th>
<th>Length of Illness (m)</th>
<th>Neurological Deficit</th>
<th>Lesion Site</th>
</tr>
</thead>
<tbody>
<tr>
<td>#1</td>
<td>51/M</td>
<td>2.5</td>
<td>M 6 SS 3 VF 6</td>
<td>TP</td>
</tr>
<tr>
<td>#2</td>
<td>67/M</td>
<td>4</td>
<td>M 6 SS 2 VF 6</td>
<td>TP, PVWM,</td>
</tr>
<tr>
<td>#3</td>
<td>68/M</td>
<td>3</td>
<td>M 6 SS 6 VF 3</td>
<td>BG</td>
</tr>
<tr>
<td>#4</td>
<td>55/M</td>
<td>12</td>
<td>M 6 SS 2 VF 6</td>
<td>TP (WM)</td>
</tr>
<tr>
<td>#5</td>
<td>45/M</td>
<td>25</td>
<td>M 6 SS 1 VF 3</td>
<td>FTPO</td>
</tr>
<tr>
<td>#6</td>
<td>69/F</td>
<td>6</td>
<td>M 6 SS 4 VF 2</td>
<td>FTP</td>
</tr>
<tr>
<td>#7</td>
<td>47/F</td>
<td>2</td>
<td>M 6 SS 3 VF 3</td>
<td>FTPO</td>
</tr>
<tr>
<td>#8</td>
<td>73/F</td>
<td>3</td>
<td>M 6 SS 2 VF 3</td>
<td>TP</td>
</tr>
</tbody>
</table>

Note. M/F = male/female. F = frontal; T = temporal; P = parietal; O = occipital; PVWM = paraventricular white matter; BG = basal ganglia. M = motor; SS = somatosensory; VF = visual half-field.

lasting about 60 min. Four procedures, briefly summarized below, were used.

1. Visual-spatial scanning. Through a dedicated PC, single digits were projected in one of 48 different positions (4 rows, each including 12 positions), on a large screen (96 degrees x 18 degrees). The distance between the patients' eyes and the screen was 100 cm. The patients' task was to press a button as quickly as possible, using the right index finger, and to name the presented digit. The procedure allowed for the presentation of sequences of varying length in different spatial positions. At the beginning of the training, sequences progressively proceeding from right to left were used. In later training stages, a digit might appear two or three positions to the left of the last presented stimulus. At the end of the training, the presented sequences included left- and right-sided stimuli in a random order. In the early stages of training, the presentation of the stimulus was accompanied by an acoustic warning signal.

2. Reading and copying. The stimuli were simple sentences and newspaper titles of realistic content. At the beginning of the treatment patients were shown simple sentences and titles printed on a single line, and were requested either to read or to copy them. As the training proceeded, more complex sentences and short stories were given. In the early stages of training, a red flashing bar was presented on the left side of the stimulus, and patients were verbally cued towards this “anchor” before starting to read or copy.

3. Copy of line drawings on a dot matrix. Stimuli consisted of two dot matrices, varying from 4 to 20 points. On the left matrix, some dots were connected by solid lines. The task was to copy this line drawing on the right dot matrix. In the early stages of training, facilitations similar to those used in the previous task were employed.

4. Figure description. Black-and-white figures of simple and realistic scenes were used. Patients were required to describe as many elements of each scene as they could. In the early stages of training, verbal cues were used.

Assessment of Visual-Spatial Hemineglect

The presence of visual-spatial hemineglect was assessed by a diagnostic battery, which included two visual-motor exploratory tasks (line and letter cancellation, for which patients used the right hand), a reading task, and a task requiring a perceptual judgment. In all tasks the center of the display was located on the mid-sagittal plane of the trunk of the patients, who were free to move their head and eyes. All patients and control subjects had a normal or corrected-to-normal vision.

1. Line cancellation (Albert, 1973). The patient's task was to cross out 21 slanted lines (2.5 cm), printed on a 42 cm x 30 cm sheet, 11 on the left-hand side, and 10 on the right-hand side. The scores were the number of omissions in the left- and right-hand sides of the sheet. Normal subjects perform this task without errors.

2. Letter cancellation (Diller & Weinberg, 1977). The patient’s task was to cross out 104 uppercase “H” letters (4 mm high), printed in six horizontal lines on a 42 cm x 30 cm sheet, 53 on the left and 51 on the right-hand side. The targets were interspersed among 208 distracters (letters). The score was the number of omissions on the left-
and right-hand sides of the sheet. The maximum number of omission errors for normal subjects is 4, and 2 is the maximum difference between errors on the two sides of the sheet (Vallar, Rusconi, Fontana, & Musici, 1994).

3. Reading (Pizzamiglio et al., 1990). Patients were asked to read aloud six sentences (mean length 8.3 words, 32.8 letters; range 5–11 words, 21–42 letters), printed in uppercase letters in the center of a 29.7 cm × 21 cm sheet. The letters were 5-mm high in three sentences, 3-mm high in the other three. The score was the number of incorrectly read sentences. Normal subjects and patients with right brain damage without hemineglect make no errors on this test. Patients with right brain damage and hemineglect make omission errors, substitution errors, or both, in the left half of the sentence.

4. Wundt-Jastrow Area Illusion Test (Massironi, Antonucci, Pizzamiglio, Vitale, & Zoccolotti, 1988). In this test each stimulus comprises two black fans of the same shape and surface. One of the two fans, however, appears longer than the other, and this illusory effect is produced either by the left or by the right sides of the fans. For each stimulus, the patient was required to communicate which fan was longer. Forty stimuli (20 with a left-sided and 20 with a right-sided illusory effect) were given in a random fixed order. Normal subjects show the expected illusory effect in all trials. Patients with left spatial hemineglect fail to show the effect when it arises in the left sides of the fans. Conversely, when the illusion is produced by the right portions of the fans, the expected effect is present. The score is the number of “expected” responses (i.e., showing the illusory effect). Patients with right brain damage and hemineglect make omission errors only on stimuli with a left-sided illusory effect. Accordingly, only these responses were considered for statistical analysis (range 0–20).

In the baseline assessment, 4 patients showed neglect in all four tasks, 3 in three tasks, and 1 patient in two tasks.

Assessment of Position Sense
The apparatus consisted of an open box (80 cm × 50 cm × 30 cm), which had a transparent plexiglass top and a wooden bottom (Vallar et al., 1993a). Transparent or black covers were placed on the plexiglass side. The four assessed positions of the patient’s forearm were drawn on the cover, as colored silhouettes (green, red, yellow, and sky blue). The arm was adducted and 30 degrees forward elevated; the forearm was 90 degrees pronated. The forearm could be moved by the examiner to four different positions: straight ahead, 30, 60, and 90 degrees adducted towards the patient’s trunk. All patients were able to point to all four positions and name the corresponding color. The box was placed on a table in front of a screen (250 cm × 300 cm), on which a dedicated PC IBM projected a set of 20 random dots (2.8 cm diameter). The stimulation area on the screen was 147 cm × 112 cm. The distance between the patients’ eyes and the screen was 100 cm. Three stimulation conditions were assessed: control (stationary luminous dots), optokinetic stimulation with a constant linear leftward movement of the dots, and optokinetic stimulation with a rightward movement of the dots. In the two latter conditions angular velocity was 45 degrees/s.

All patients showed the normal optokinetic nystagmus response, which was assessed by modified Frenzel glasses, prior to the experimental study. A Frenzel lens was applied on the left eye, while subjects were looking at the moving stimuli with their right eye.

During practice, the patient’s forearm was placed on the bottom of the box and the transparent cover was used. In each trial the starting point was the straight ahead position of the forearm, which was repeatedly moved towards and away from the trunk, before the intended position was reached. During training, patients could see the position of their forearm under the transparent screen. They received instructions to look at the screen in front of them while the examiner moved their forearm to the intended position, and to communicate the perceived position of the forearm, by pointing to the corresponding silhouette on the transparent display, by naming its color, or both. Three stimulation blocks were given in the following order: control (stationary dots), dots with a leftward movement, and dots with a rightward movement. In each block 12 trials were given (3 per position). The intertrial interval was about 5 s.

The experimental session differed from practice in that the box cover was black, preventing patients from viewing their forearm. The four tested forearm positions and their colored drawings on the black cover were identical to those used in the practice session. Four blocks were given: control-1 (stationary dots), stimulation with a leftward direction of the movement, stimulation with a rightward direction of the movement, and control-2 (stationary dots). Each block comprised 40 trials (10 for each position) in a random order. In each block, the stimulation started 40 s before the first trial and was stopped after the completion of the last trial. Patients did not look at the screen, while they provided their response looking at the horizontal box.
In each block the score was the number of correct responses (range 0–40), that is, a reported position of the forearm corresponding to the actual position. In blocks 1–4, the sequences of optokinetic stimulation (leftwards, rightwards) were counterbalanced across subjects. For the purpose of statistical analysis, the scores of the two control conditions (before and after stimulation) were averaged.

In patients with lesions in the right hemisphere and left visual-spatial hemineglect, the deficit of position sense is much more severe in the contralesional arm, as compared to the right arm, in which a slight, though significant, impairment is present (Vallar et al., 1993a; Vallar et al., 1995). In the present series, the severity of the deficit in the left forearm (see Fig. 2, control condition, before treatment) was comparable to that observed in our previous studies (average accuracy about 50%) (Vallar et al., 1993a; Vallar et al., 1995). By contrast, patients with damage to the left or to the right hemisphere, but with no evidence of hemineglect were much more accurate: Their error rate in the contralesional arm was about 10% (Vallar et al., 1993a; Vallar et al., 1995). The present baseline data confirm therefore the association between severe deficits of position sense and visual-spatial hemineglect (Vallar et al., 1993a; Vallar et al., 1995; Willanger et al., 1981).

Finally, because the present experiment aimed at assessing the effects of a rehabilitation treatment for visual-spatial hemineglect on position sense disorders, the study was confined to the more severely impaired left forearm. The deficit of position sense in the right ipsilesional forearm was assessed in the pre-treatment assessment only, without optokinetic stimulation. The patients’ average correct responses were 36.47 out of 40 trials (91.2%), in line with previous findings (Vallar et al., 1993a; Vallar et al., 1995).

Procedure and Statistical Analysis
The assessments of extra-personal visual-spatial hemineglect and of the deficit of position sense were performed before the beginning of the rehabilitation treatment, and after its completion, 2 months later.

The patients’ scores in the four tests assessing extra-personal visual neglect were transformed in z scores, on the basis of the performances of 118 patients with damage to the right hemisphere and visual-spatial hemineglect (Pantano et al., 1992). The scores obtained by the patients on each test, before and after the treatment, were analyzed by paired t tests. The patients’ scores in the task assessing deficits of position sense were treated by an analysis of variance with two within-subject factors: Rehabilitation (before and after), and stimulation condition (control, optokinetic stimulation with ipsilateral/contralateral direction of the movement).

RESULTS
As shown in Figure 1, the patients’ scores on the four tasks assessing extra-personal visual-spatial hemineglect improved after the treatment. Paired t tests showed significant differences in the letter cancellation ($t = 3.75, df = 7, p < 0.01$) and reading ($t = 3.38, df = 7, p < 0.05$) tests, and a trend in the line cancellation ($t = 2.16, df = 7, p = 0.068$) and the Wundt-Jastrow illusion ($t = 1.91, df = 7, p < 0.098$) tasks. Table 2 shows that the rehabilitation treatment improved the deficit in all patients in whom the first assessment had revealed evidence of hemineglect.

Figure 2 shows the patients’ performance in the position sense task. An analysis of variance showed a significant main effect for stimulation ($F = 7.854; df = 2, 14; p < 0.01$), but neither the main effect for rehabilitation ($F = 2.71; df = 1, 7$), nor the interaction ($F = 1.66; df = 2, 14$) were significant. Duncan’s tests showed that the contralesional stimulation condition differed from both the control and the ipsilesional stimulation conditions ($p < 0.01$). The rehabilitation treatment neither improved the deficit of position sense nor modified the effects of optokinetic stimulation. Stimulation with a contralesional direction of the movement improved the disorder of position sense, whereas the ipsilesional direction had minor negative effects.

Finally, the correlation between the effects of the rehabilitation treatment on the patients’ performances on tests assessing visual-spatial hemineglect and the deficit of position sense was computed. The differences between the average z scores in the four tasks assessing hemineglect and in the test assessing position sense (control condition) before and after the treatment were ranked. The rank order correlation was not significant ($r_s = .20$).
Table 2. Number of Patients who Showed Left Visual-Spatial Hemineglect before Rehabilitation, and Improvement after the Treatment, by Task Assessing Extra-Personal Visual-Spatial Hemineglect.

<table>
<thead>
<tr>
<th>Cancellation</th>
<th>Sentence Reading</th>
<th>Wundt-Jastrow Illusion</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Letter</td>
<td>Line</td>
</tr>
<tr>
<td>Neglect</td>
<td>8/8</td>
<td>6/8</td>
</tr>
<tr>
<td>Improved</td>
<td>8</td>
<td>6</td>
</tr>
</tbody>
</table>

DISCUSSION

The specific rehabilitation program used in this study, focusing on the exploration of visual extra-personal space, improved left visual-spatial hemineglect in all 8 patients, replicating previous observations (Antonucci et al., 1995; Pizzamiglio et al., 1992). The treatment, however, neither affected the severity of the deficit of position sense in the left arm, nor modified the sensitivity of the position sense disorder to optokinetic stimulation. In line with previous findings (Vallar et al., 1993a; Vallar et al., 1995), optokinetic stimulation with a leftward direction of the movement of the luminous dots temporarily improved the disorder, although stimulation with a rightward direction was ineffective. The presence, and persistence after the rehabilitation procedure, of the direction-specific effects of optokinetic stimulation, which temporarily improves also visual-spatial hemineglect (Karnath, 1996; Pizzamiglio et al., 1990; Vallar, Guariglia, & Rusconi, 1997), indicates that the position sense deficit of the present group of patients with damage to the right cerebral hemisphere had a neglect-related component. This, however, unlike extra-personal visual-spatial hemineglect, was not affected by

Fig. 1. Average severity (z scores, SD) of left visual-spatial hemineglect in 8 patients with lesions in the right hemisphere, before and after the rehabilitation treatment (PRE/POST). Tests: line and letter cancellation, Wundi-Jastrow illusion (W-J), reading.
Fig. 2. Position sense in the left forearm before and after the rehabilitation treatment (PRE/POST): average correct responses in the baseline condition (CONTROL), and during optokinetic stimulation with a contralesional, leftward (CONTRA), and ipsilesional, rightward (IPSI), direction of the movement of the luminous dots.

The present findings support this view from the perspective of unilateral neglect, distinguishing the spatial co-ordinate systems concerned with visual-spatial hemineglect and the deficit of position sense reflect the impairment of a unitary spatial representational system, concerned with both objects in extra-personal space, and the position of body segments.

The view that discrete spatial systems are involved in the internal representation of the position of body segments and of the environment surrounding the subject is consistent with a number of independent observations. Flanders and Soechting (1995), through the analysis of errors in reaching and grasping movements of normal subjects, suggest the existence of spatial reference frames "fixed to the arm", which may be distinguished from frames "fixed in space" (head- or body-centered). The existence of spatial frames of reference related to the spatial position of body parts is also supported by neurophysiological observations in the monkey: During the execution of arm-reaching movements, cortical activity in the motor and in the premotor cortices varies with the initial position of the arm (Caminiti, Johnson, Galli, Ferraina, & Burnod, 1991; Caminiti, Johnson, & Urbano, 1990). Finally, Graziano and Gross (1995, 1993, 1994) have described in a number of cerebral regions (parietal area 7b, premotor area 6, putamen) cells with bi-modal, visual-tactile properties. For these cells, the visual receptive field matches the location of the tactile field and is confined in depth to a region near to the animal. In many such neurons the visual receptive field moves along the tactile receptive field, when the arm or the hand is placed in different locations. On the basis of these findings, they suggest that visual stimuli may be coded in arm-centered co-ordinates, and, more generally, the existence of body-part-centered representations, which are closely related to the near visual space.
with the position of body parts, such as the forearm, from those involved with the egocentric representation of objects in extra-personal space. In line with this conclusion, patients with damage to the right cerebral hemisphere may show neglect for the left side of the body, but not for left-sided objects and vice-versa (Bisiach, Perani, Vallar, & Berti, 1986; Guariglia & Antonucci, 1992). These “personal” and “extra-personal” discrete co-ordinate systems, however, share the property of being modulated by a number of afferent sensory sources (vestibular (Cappa, Sterzi, Vallar, & Bisiach, 1987; Vallar et al., 1993b), visual (Karnath, 1996; Pizzamiglio et al., 1990; Vallar et al., 1993a; Vallar et al., 1995), proprioceptive/somatosensory (Karnath, 1994; Karnath, Christ, & Hartje, 1993). In the different manifestations of spatial hemineglect (defective perceptual awareness and visual-motor exploration of extra-personal space, the neglect-related components of hemianaesthesia and of the deficit of position sense) a basic disorder may be a distortion of a given co-ordinate system towards the side of the lesion. This leads to an impoverished representation of the contralateral side, which may be restored, temporarily and in part, by direction-specific sensory stimulations, such as the optokinetic input used in the present study (Vallar et al., 1993a; Vallar et al., 1997).

The differential effects of optokinetic stimulation on the one hand, which may temporarily improve or worsen virtually all of the investigated clinical manifestations of the neglect syndrome (review in Vallar et al., 1997), and of the present rehabilitation treatment on the other, which produces a persistent improvement confined to visual-spatial hemineglect, may reflect different compensatory mechanisms set up by the two procedures. Direction-specific optokinetic stimulation, as other sensory inputs, may affect higher order (e.g., egocentric) co-ordinates across discrete spatial representations, modulating the ipsilesional distortion produced by the cerebral lesion. The rehabilitation treatment, by contrast, may improve the exploration of a specific neglected sector of space (in the present experiment, visual extra-personal space), but this does not extend to other spatial representations.

A final practical implication of the dissociation between representations for the spatial positions of body segments and visual extra-personal space concerns the design of rehabilitation programs in patients with hemineglect. The present observation that the effects of a training procedure concerning the defective exploration of visual extra-personal space do not extend to the neglect-related deficits of position sense indicates that the latter requires specific rehabilitation programs.

REFERENCES


