

Left size distortion (hyperschematia) after right brain damage

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Abstract—Objective: To quantitate a size distortion involving the side of space contralateral to the lesion (contralesional) in two right-brain-damaged patients. **Methods:** We studied two right-brain-damaged patients with lesions sparing the occipital lobe and a mild left neglect on target cancellation or line bisection. The lesions involved the temporoparietal region (Patient 1) and the basal ganglia and the insula (Patients 1 and 2). Patients were given drawing tasks and tasks requiring perceptual and visuomotor judgments of horizontal extent. **Results:** In drawing objects such as a daisy both from memory and by copying, patients exhibited a disproportionate enlargement of the left-hand side of objects and added more left-sided petals to the drawn daisy. This pathologic behavior persisted when the patients were blindfolded and was likely to reflect a perceptual, rather than premotor, size distortion. In a task requiring the perceptual matching of two rectangles, patients underestimated the left-sided stimulus. In a visuomotor task requiring the reproduction of the horizontal extent of a segment, patients exhibited a hyperextension, when a leftward movement was required. **Conclusions:** We showed a disordered representation of extrapersonal space, possibly involving a contralesional relaxation of the spatial medium. The deficit does not arise at the level of retinotopic coordinate frames and is independent of unilateral spatial neglect.

NEUROLOGY 2006;67:1801–1808

Lesions of the right cerebral hemisphere may affect drawing tasks. Some deficits are constructional,^{1,2} whereas others are closely associated with left unilateral spatial neglect. Right-brain-damaged patients with left neglect typically omit left-sided details in drawing from memory and in copying^{3,4} and may show graphic perseveration in cancellation tasks.^{5,6} This perseveration behavior in the side of space ipsilateral to the side of the lesion (ipsilesional) ranges from the repeated canceling out of targets to the addition of new targets and more complex and gratuitous graphic productions. Much less frequently, patients with left neglect exhibit a left-sided expansion of the drawing, contralateral to the side of the lesion (contralesional), when required to draw symmetric objects from memory.^{7,8} This leftward contralesional expansion of the left-hand side of symmetric drawings is somewhat paradoxical, associated with the omissions of left-sided contralesional details, committed by patients with left neglect in cancellation tasks.³ Ipsilesional perseveration and contralesional expansion in drawing tasks may be considered productive manifestations of neglect, which are sometimes associated with the more frequent defective manifestations, such as left-sided omissions in drawing tasks.⁴

Available evidence about the disproportionate expansion of the contralesional side of objects in drawing tasks is limited and concerns clinical observations in patients with left neglect⁷ or corollary findings in studies assessing other aspects of the neglect syndrome.⁸ One patient drew a daisy from memory with more and greater petals on the left side.⁷ Another patient drew a clock with the left-sided hours displaced to the left, outside the clock face, occupying more space leftward. In this patient transcutaneous nervous electrical stimulation, which improved left neglect in other tasks, abolished the leftward expansion.⁸ We report a quantitative investigation of the performance of two right-brain-damaged patients with a disproportionate left-sided expansion in drawing tasks.

Case reports. *Patient 1.* A 61-year-old right-handed man, with 9 years of schooling, who had been working as an electrician, with no previous neurologic or psychiatric disorders and a history of hypertension, was admitted to a neurologic unit for the sudden onset of a left-sided weakness. A neurologic examination showed a left hemiplegia, with increased deep tendon reflexes, an extensor plantar response, and a left hemianesthesia. No head and gaze deviation toward the side of the lesion was noted. No visual field deficits were detected on confrontation. The patient was alert and cooperative and showed left spatial neglect on a clinical bed-side assessment. Three months after stroke onset, the patient showed

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Supported in part by Miur Prin grants 2003 and 2005, a FAR Grant 2006 (Dr. Vallar), and an SNLF Grant (Dr. Michel).

Disclosure: The authors report no conflicts of interest.

Presented in part to the Joint Meeting of the Société de Neuropsychologie de Langue Française and the Società Italiana di Neuropsicologia (Paris, October 17–18, 2002).

Received December 29, 2005. Accepted in final form July 28, 2006.

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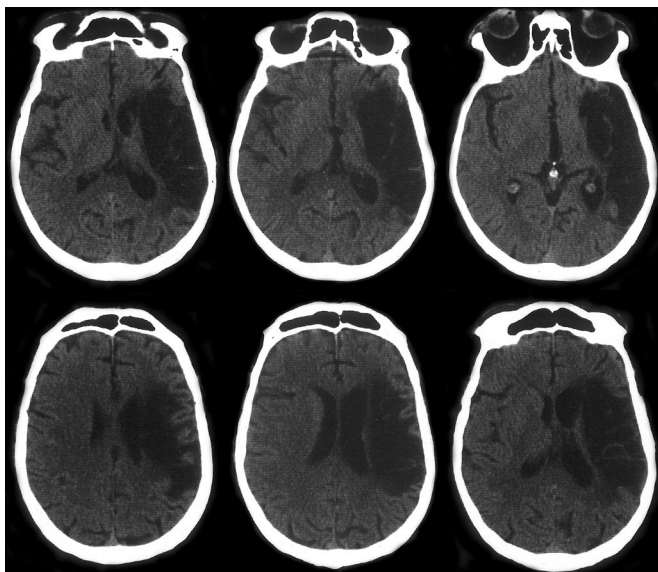


Figure 1. Right hemispheric lesion in Patient 1 (CT scan).

a severe left spastic hemiparesis and a severe left somatosensory deficit. The visual fields were normal, as assessed by Goldmann perimetry. Visual acuity was corrected to normal (10/10) with lenses for the two eyes. No auditory or visual extinction to double simultaneous stimulation was noted. The patient was aware of his motor and somatosensory deficits.

The patient scored normally on a line cancellation test, crossing out 40 of 40 lines,⁹ and on a star cancellation test, scoring 53 of 54 targets, with one omission in the center of the display.¹⁰ The patient showed no neglect on a drawing copy task.¹¹ In drawing from memory, however, the patient produced some putatively symmetric objects (a house, a fir tree, a Christmas fir tree, a butterfly, a daisy) disproportionately larger on the left side. This behavior was particularly evident in the case of the daisy.

Brain CT (figure 1), performed 18 months after stroke onset, showed an extensive single lesion in the vascular territory of the right middle cerebral artery. The lesion involved the temporoparietal region, the internal capsule, the head of the caudate nucleus, the putamen, the insula, and the corona radiata, particularly in the anterior part. Angiography showed a complete obstruction of the right middle cerebral artery, before its division into different branches.

Patient 2. A 45-year-old right-handed woman, with 16 years of schooling, who had been working as a bookkeeper and with no history of neurologic or psychiatric disorders, was admitted to a neurologic unit owing to the sudden onset of a left-sided motor weakness. A neurologic examination showed a left hemiplegia, a rightward deviation of the head and eyes, a moderate dysarthria, and denial of illness. In a few days, the patient's deficit rapidly recovered, with a neurologic exam showing a mild facial paresis and weakness of the left hand. A general apathy and emotional flattening and a reduced spontaneous utilization of the left limbs (motor neglect) were noted.¹² Three weeks after stroke onset, the patient showed a reduced dexterity in the left hand. No somatosensory and visual field deficits were present. No auditory, visual, or somatosensory extinction was found. The patient showed a mild left unilateral neglect in a letter cancellation task,¹³ omitting four targets in the far left-hand side of the display and no right-sided targets. No evidence of neglect was found in a line cancellation task,⁹ in a bell cancellation task,¹⁴ in copying the Rey figure,¹⁵ in a drawing copy task,¹¹ and in a representational task (map of France).¹⁶ In drawing from memory, it was noted that the patient produced some putatively symmetric objects (a star, a heart, a butterfly) larger on the left side.

MRI exam (figure 2), performed 3 months post onset, showed a deep lesion involving the head of the caudate nucleus, the putamen, the insula, the internal capsule, and the frontoparietal paraventricular white matter.

Both patients gave their informed consent prior to the study.

Six right-handed neurologically unimpaired subjects (three

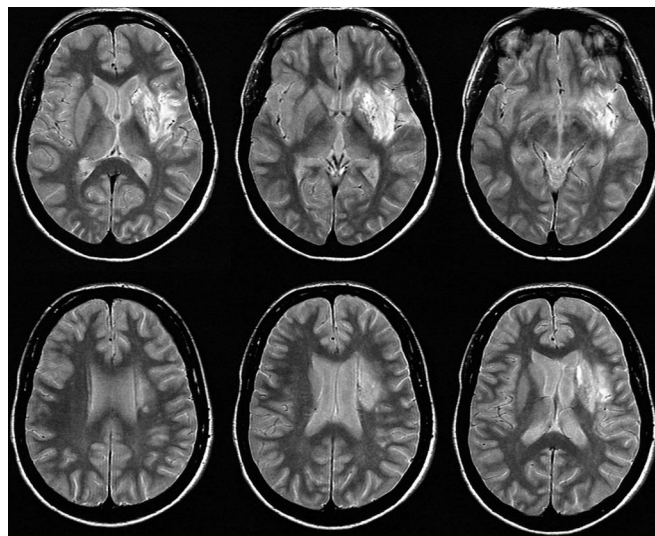


Figure 2. Right hemispheric lesion in Patient 2 (MRI axial T2-weighted images).

men and three women, mean age 54.8 years [range 34 to 78 years], mean years of schooling 12 [range 9 to 18 years]) participated in the study as control subjects.

Results. Line bisection. The test devised by Schenkenberg et al.¹⁷ was used. The stimuli included 18 lines. Eighteen lines were organized in three sets of six lines, so that one set lay primarily on the left side of the page, one in the center, and one on the right side. Each set contained lines of 100, 120, 140, 160, 180, and 200 mm. The lines were organized so that the test was balanced with regard to line length from top to bottom. Two 150-mm lines, placed at the top and bottom (center), were used in communicating instructions to the subject and were not included in data analysis. Subjects received instructions to mark the center of each line with a soft pen, without skipping any. Patients and control subjects used the right hand, namely, the hand ipsilateral to the side of the lesion in the two brain-damaged patients. The length of the left side of the line (i.e., from the left end of the line to the subject's mark) was measured to the nearest millimeter. That measurement was converted to a standardized score (percentage deviation), using the following formula: measured left half – objective half/objective half × 100. This transformation yields positive numbers for marks placed to the right of the center and negative numbers for marks placed to the left of the center.

Patient 1 showed a +4.0% deviation and Patient 2 a –2.0% deviation. The average percentage deviation of control subjects was –0.7 (SD ±2.46, range –4.3 to +2.9). The patients' performances were compared with the control data by *t* tests,^{18,19} looking for a rightward bias in bisection, associated with the right-hemispheric lesion. A trend toward significance ($t = 1.773$, one-tailed $p = 0.068$) was found for Patient 1 and no difference for Patient 2 ($t = -0.487$, one-tailed $p = 0.323$). The mild rightward bias shown by Patient 1 is consistent with the clinical evidence of a left spatial neglect in the early stages of his disease. The performance of Patient 2 was well within the normal range, with, if anything, a leftward bias.

Drawing from memory. Subjects were required to draw from memory a daisy, on a 21 × 29.7-cm sheet of

paper, placed in front of them, with the center of the sheet aligned with the midsagittal plane of the patients' body. No model was provided, and no further specific instructions were given. To measure the area and the number of petals of the left and of the right sides, each drawn daisy was divided into two sides by a vertical line passing through the center of its pistil. Petals divided into approximately equal parts by the vertical line were not considered in counting the number of drawn petals. Occasionally, patients added leaves, which were not considered for the analyses. The areas of the two sides of the drawing were computed by a Leica imaging system and a Quantimet 500 software. For the left- and right-sided areas of each drawing, a laterality index (LI) score was computed: $(\text{left-sided area} - \text{right-sided area} / \text{left-sided area} + \text{right-sided area}) \times 100$. A positive value of this LI indicates a greater left-sided area and a negative LI a greater right-sided area. A similar LI was computed for the number of petals (PLI). Each patient was required to draw six daisies, as well as the control subjects. The two patients showed no difficulties in executing the task and drew the pistil and the right-sided petals first. Control subjects, too, drew the pistil first, but did not show any definite pattern as to the petals.

The role of input (perceptual) vs output (premotor) factors in bringing about the pathologic enlargement of the left side of drawings was assessed by the epidiascope method.²⁰ This procedure, originally devised to tease apart these component factors in patients with unilateral spatial neglect,²¹ decouples the direction of the hand movement from the subject's visual control of the display. Patients with a perceptual disorder would show a left-sided deficit (omissions in a cancellation task, a disproportionate enlargement of the left-hand side of the drawing in the current investigation) with reference to their field of vision (normal vs mirror-reversed), independent of the direction of the movement of the arm and hand. Patients with a premotor disorder would exhibit a left-sided deficit, with reference to the midsagittal plane of the body, independent of the normal or mirror-reversed field of vision.

In Patient 1 the role of visual control was assessed by a blindfold condition (three trials were given). In this study, control data were collected from six neurologically unimpaired subjects (three men and three women; mean age 54.5 years, range 30 to 81 years), with three subjects being different from those participating in the other experiments. Each control subject was given six trials.

Figure 3A shows examples of drawing from memory by the two patients. Figure 4 shows the patients' mean LI and PLI scores. The positive mean LIs of both patients, well outside the range of control subjects, indicate that they drew the left-hand side of the daisy larger than the right-hand side. Control subjects, by contrast, drew a smaller left-hand side of the daisy, different from zero (mean LI: -4.87 , $SD \pm 1.34$, $t_5 = -8.88$, $p < 0.001$). The patients' scores were compared with the control data by t tests.^{18,19} For the LI score, differences were found for both Patient 1 ($t = 22.71$, $p < 0.001$), and Patient 2 ($t = 9.24$, $p < 0.001$). All daisy drawings by Patient 1 had a positive LI score (range $+16.77$ to $+44.18$). In Patient 2, all drawn daisies but one item had a positive LI score (range -13.28 to $+35.10$). In sum, in patients, 11 of 12 (91.6%) drawn daisies had a positive LI score; in the six control subjects, only

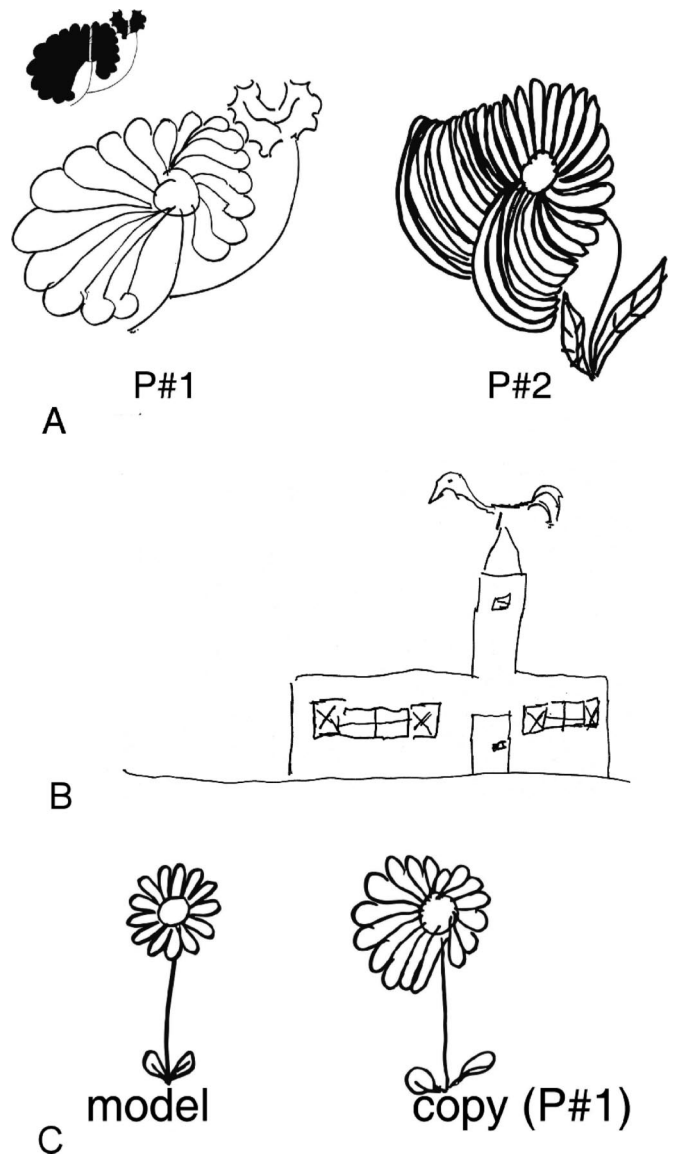


Figure 3. Examples of drawing. (A) From memory: Patients 1 and 2 (daisy). The top left black silhouette with a white space between the two sides of each drawing shows the measured areas in the drawn daisies. Patient 1 drew also a greater left-hand side of a leaf located to the right of the daisy. (B) From memory: Patient 1 (church). Within the church, the windows show a greater left-hand side. (C) By copy: Patient 1 (daisy).

6 of 36 (16.6%) drawn daisies scored positively (χ^2 Yates corrected = 18.98, $df = 1$, $p < 0.0001$).

On average, both patients drew more petals on the left side. The PLI of Patient 1 was outside the controls' range; the PLI score of Patient 2 was just within the positive limit of the controls' range. The patients' PLI scores were compared with the control data.^{18,19} A trend toward significance ($t = 2.41$, $p = 0.067$) was found for Patient 1 and not for Patient 2 ($t = 1.42$; NS). In control subjects, the negative PLI score, indicating less left-sided drawn petals, did not differ from zero (mean PLI: -2.39 , $SD \pm 4.03$, $t_5 = -1.45$, NS).

In the epidiascope condition, both patients drew a larger left-hand side of the daisy, with reference to their

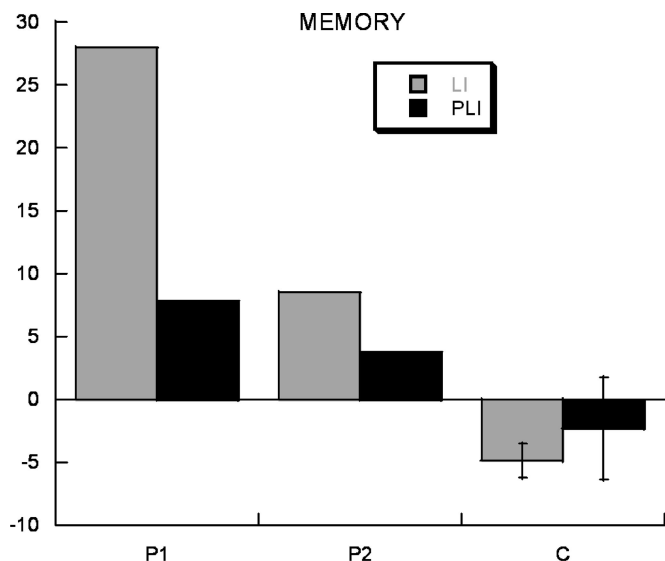


Figure 4. Drawing a daisy from memory. Mean laterality indexes (LIs) for drawn areas and mean LI for number of petals (PLIs) in Patients 1 and 2 and six control subjects (mean and range).

field of vision. Indeed, the direction of the hand movement was preferentially oriented toward the right side. The LIs, computed on the seen drawing, were +24.4 for Patient 1 and +8.7 for Patient 2. Values of LI for number of petals were, respectively, +22.2 for Patient 1 and +11 for Patient 2.

In the blindfold condition, without visual control, Patient 1 drew a left-hand side of the daisy much larger than the right-hand side, on all three trials: The LI score was +56.0 (range +32.3 to +79.2). The average LI score of six control subjects was -8.72 ($SD \pm 4.71$; range -2.66 to -14.85). The LI score of the patient was compared with the control data by a *t* test,^{18,19} showing a difference ($t = 12.72$, $p < 0.001$). The PLI score was +40.0 (+14.2 to +53.8). The average PLI score of six control subjects was -8.19 ($SD \pm 3.62$, range -4.32 to -14.10). A *t* test comparing the PLI score of the patient with the control data^{18,19} showed a difference ($t = 12.32$, $p < 0.001$).

Patients showed a similar left-hand-side enlargement with other drawings from memory: a church (LI = +25.0) (figure 3B), a face (LI = +11.8), an apple tree (LI = +3.3), and a tree (LI = 7.4) for Patient 1; a butterfly (LI = +15) and a tree (LI = +42) for Patient 2.

Copying. This test differed from the previous one in that a symmetric daisy model was provided. The model was printed in the center of a 21×29.7 -cm sheet. Six trials were given. The data were analyzed as in the drawing from memory study. The same subjects who took part in the previous experiment provided control data. Patient 2 was not available for testing. Figure 3C shows an example of copy drawing by Patient 1. Figure 5 shows the LI and PLI scores that were outside the controls' range. The patient drew a larger left-hand side of the model and more left-hand petals. The patient's performances were compared with the control data by *t* tests.^{18,19} A difference was found for the LI score ($t = 8.911$, $p < 0.001$) and not for the PLI score ($t = 1.351$, NS). Control subjects drew a smaller left-hand side of the daisy (mean (LI: -4.18 , $SD \pm 2.72$),

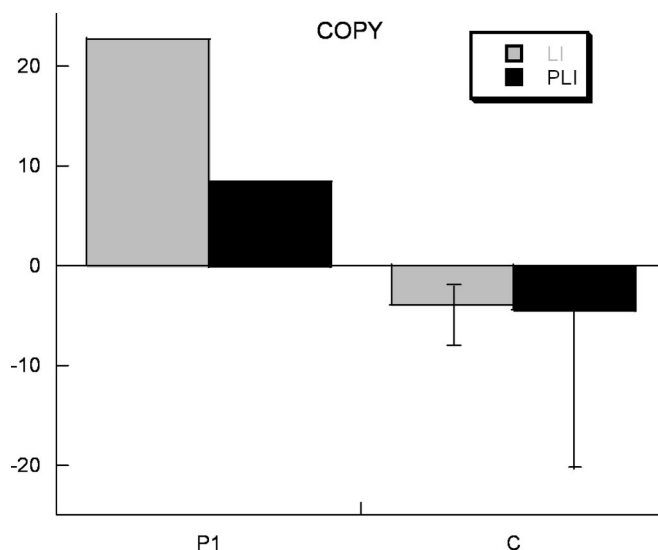


Figure 5. Drawing a daisy by copying. Mean laterality index (LI) for drawn areas and mean LI for number of petals (PLI) in Patient 1 and six control subjects (mean and range).

different from zero ($t_5 = -3.76$, $p = 0.013$). The PLI score (mean PLI: -4.41 , $SD \pm 8.15$) did not differ from zero ($t_5 = -1.32$, NS). In the patient, 5 of 6 LI (range -9.12 to $+41.47$) scores (83%) were positive, whereas in control subjects, the positive scores were 11 of 36 (30.5%) (χ^2 Yates corrected = 4.04, $df = 1$, $p < 0.05$).

Patients showed a similar left-hand-side enlargement with other drawings by copy: a tree (LI = +7.4) for Patient 1 and a butterfly (LI = +10.8) and a tree (LI = +39.5) for Patient 2.

Perceptual matching task. The stimuli were pairs of black rectangles 15 mm high, with a 6-cm empty space between them. Twenty-five pairs of rectangles were presented in a pseudorandom series to measure the point of subjective equality between patterns placed in the left and right visual half-spaces. The distance between the right side of the left-sided rectangle and the left side of the right-sided rectangle was 8 cm. The center of this distance was aligned with the midsagittal plane of the subjects' body. In each pair, the length of one rectangle was fixed (8 cm), and the length of the other varied from 6.4 to 9.6 cm, in four 8-mm steps. In five trials, the two rectangles were equal in length (8 cm). In 10 trials, the right-sided rectangle was longer than the left-sided segment, and in 10 trials vice versa. The subject's task on each trial was to report verbally which was the longer out of the two rectangles. Each error on a given trial was scored a value of $n \pm 1$, where n was the number of steps by which the patterns' lengths differed on that trial. Rightward errors (i.e., the right-sided rectangle judged as longer, when the left-sided rectangle was longer) were given a positive score. Leftward errors (i.e., the left-sided rectangle judged as longer, when the right-sided rectangle was longer) were given a negative score. With use of this scoring method,²² an identical pair of stimuli ($n = 0$) yielded a score of either +1 (rightward error) or -1 (leftward error). The larger was the difference in length between the two rectangles, the greater the error score.

Patient 1 scored +1.31, and Patient 2 scored +1.19. The

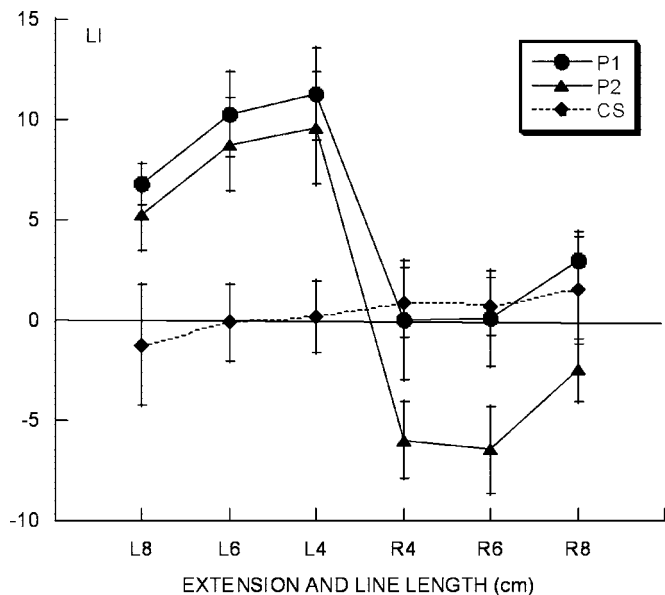


Figure 6. Line extension task. Mean (SE) laterality indexes (LIs) of Patients 1 and 2 and six control subjects for rightward and leftward (L, R) extended lines (line length: 4, 6, and 8 cm).

mean score of the control group was $+0.905$ (SD ± 0.11 , $+0.81$ to $+1.07$). The patients' scores were outside the controls' range, suggesting a relative perceptual underestimation of the lateral extent of the left-sided rectangle compared with the right-sided rectangle. The patients' performances were compared against the control data by *t* tests.^{18,19} A difference was found for Patient 1 ($t = 3.41$, $p = 0.019$). The difference approached the 0.05 level for Patient 2 ($t = 2.40$, $p = 0.067$).

Line extension task. The subject's task was to reproduce the length of a horizontal line in two conditions. In the rightward movement condition, the right end of the line was aligned with the midsagittal plane of the body, and the patient reproduced the perceived length of the segment with a rightward extension. In the leftward movement condition, the left end of the line was aligned with the midsagittal plane of the body, and the patient made a leftward extension. The stimuli were horizontal black lines, 1 mm in width, with three line lengths (4, 6, and 8 cm). In each movement condition, in Patient 1, 18 lines were presented, 6 per each length, in a random fixed order. In Patient 2, 24 lines were presented. The length of the segment drawn by each subject on each trial was measured to the nearest millimeter. For the leftward extension of each line drawing, an LI score was computed: $\text{leftward extended length} - \text{length of the right-sided line} / \text{leftward extended length} + \text{length of the right-sided line} \times 100$. For the rightward extension of each line drawing, the LI was as follows: $\text{rightward extended length} - \text{length of the left-sided line} / \text{rightward extended length} + \text{length of the left-sided line} \times 100$. A positive value of this LI indicates overextension and a negative value hypoextension. The mean LIs were calculated for the two directions of extension (rightward vs leftward) and for the three line lengths (4, 6, and 8 cm).

Figure 6 shows the average LIs of the two patients and of the six control subjects. Both patients showed a leftward overextension. Patient 1 had a preserved performance in

the rightward extension condition, and Patient 2 showed a rightward hypoextension. Control subjects showed no major left-right differences and, if anything, some rightward greater extension. The patients' performances were analyzed by two Kruskal-Wallis one-way analyses of variance by ranks. In Patient 1, a difference among conditions was found ($KW_5 = 16.21$, $p = 0.063$). Multiple comparisons showed differences for all leftward vs rightward extensions, for leftward 8 vs 6 cm, and 8 vs 4 cm extensions, and for rightward 8 vs 6 cm extensions ($p < 0.05$). The differences between leftward 6 vs 4 cm extensions, rightward 8 vs 4 cm and 6 vs 4 cm extensions were not significant.

In Patient 2, a difference among conditions was found ($KW_5 = 28.59$, $p < 0.0001$). Multiple comparisons showed differences for all leftward vs rightward extensions and for both leftward and rightward 8 vs 6 cm and 8 vs 4 cm extensions ($p < 0.05$). The differences for leftward and rightward 6 vs 4 cm extensions were not significant.

In neurologically unimpaired control participants, a Friedman two-way analysis of variance by ranks showed no differences between the six conditions ($\chi^2_5 = 2.70$, NS).

Finally, to further assess whether the performances of the two patients differed from those of control subjects, the overall LIs were computed. In Patient 1, the LIs were $+9.4$ for leftward extension and $+1.02$ for rightward extension. In Patient #2, the LIs were $+7.9$ and -4.97 . In the control group, the mean LI was $+0.08$ (SD ± 2.4) for leftward extension and $+1.9$ (SD ± 1.9) for rightward extension. The performance of both patients showed leftward overextension. In rightward extension, the performance of Patient 1 was within the normal range, whereas Patient 2 exhibited hypoextension. The patients' performances were compared with the control data by *t* tests.^{18,19} Differences were found for both patients for leftward extension (Patient 1: $t = 3.59$, $p = 0.016$; Patient 2: $t = 3.02$, $p < 0.05$), but not for rightward extension (Patient 1: $t = 0.34$, NS; Patient 2: $t = 1.50$, NS).

Discussion. Two right-brain-damaged patients showed a systematic enlargement of the left-hand side of objects drawn from memory and by copying (Patient 1). This left-sided distortion of size in drawing tasks was influenced neither by the direction of the movement of the hand nor by visual control (Patient 1). Accordingly, the size distortion does not reflect a disorder of motor programming and action execution in the left side of space, as it takes place in the left field of vision, independent of the direction of the movement of the upper limb and the hand, manipulated through a mirror-reversed vision of action (Patients 1 and 2). Second, in Patient 1, the deficit was not confined to drawing objects under visual control, extending instead to internally generated (possibly visual and proprioceptive) representations.

This pathologic expansion of the left side of objects in drawing resembles some aspects of a rare disorder of visual perception, termed "hemimicropsia," characterized by a reduction of the perceived size of objects in one visual half-field.^{23,24} Patients with hemimicropsia typically see objects in the contralateral hemifield smaller in size compared with objects in the ipsilesional hemifield. Hemimicropsia

patients also tend to compensate for the deficit: They draw the contralesional side of objects larger than the ipsilesional side and correct symmetric patterns, making the contralesional side larger. In a size comparison task, these patients underestimate the size of the contralesional object. Hemimicropsia is considered a disorder of size perception arising in the visual fields, in retinotopic reference frames. In line with this view, the responsible lesion most frequently involves the visual association cortex (Brodmann areas 18 and 19).²³⁻²⁵ Contralesional hemimicropsia is usually not associated with unilateral spatial neglect.^{23,24} In one study²³ 2 days after onset, Patient 1 showed a mild left neglect (not documented psychometrically), which had probably receded 1 week later, when left hemimicropsia was assessed.

The performance of the current patients differs in important respects from the pattern of hemimicropsia. First, the size distortion is still present without visual control (Patient 1). This indicates that the deficit extends to internally generated visual images and is not confined to the visual perception of objects. Hemimicropsic Patient PS²⁴ was not tested without visual control, but “on specific questioning she did not refer to distorted size during dreaming or when she imagined objects with eyes closed.” Second, both patients, in addition to drawing objects larger on the left-hand side, add left-sided material, such as petals in daisy drawing; the effect is more pronounced in Patient 1. Third, both patients reported here are entirely unaware of their deficit in drawing, like patients with unilateral spatial neglect.²⁶ By contrast, patients with hemimicropsia are typically aware of the deficit in size perception, reporting that objects in the contralateral hemifield appear “smaller,” or “somewhat shrunken and compressed,”²³ or that “everything on the left side appeared distorted in its size.”²⁴

In the current patients, the distortion observed in the drawing and size comparison tasks cannot be explained in terms of a general unilateral spatial neglect for the left side of space.^{3,27} In the reported patients, evidence of a mild left spatial neglect was found. Patient 1 showed a small rightward error in line bisection, but no neglect in cancellation and drawing tasks; Patient 2 omitted a few letters in a cancellation task in the far left side, but showed no neglect on line bisection. Patients with left neglect may be selectively impaired in target cancellation and line bisection, suggesting that these tasks assess different components of the neglect syndrome.^{28,29} The current finding that the size distortion deficit was associated with a mild impairment, in different patients, in only one of these two tasks, suggests that neglect and the size distortion reported here are independent, though co-occurring, spatial disorders. Furthermore, in the main tasks that revealed size distortion, namely, drawing from memory and by copy, both patients showed, if anything, an increase of drawing activity in the contralesional side, as indexed by the number of drawn petals. By contrast, in

drawing tasks, right-brain-damaged patients with left neglect typically omit left-sided details.^{3,27} This dissociation between neglect and size distortion suggests that the spatial processes involved may differ in important respects. Cancellation tasks typically require a step-by-step exploratory activity, which is unlikely to be affected by a deficit primarily concerning the represented size of objects. Line bisection does not involve the reproduction by drawing of the bidimensional sizes of the two sides of a supposedly symmetric object (e.g., as in copying, or drawing by memory, a daisy) or of the length of a segment, as in the line extension task. Also, bisection does not require the explicit comparison of the sizes of two objects, as in the perceptual matching task.

A further difference between unilateral spatial neglect and the current size distortion concerns perseveration behavior. Some right-brain-damaged patients, particularly when the lesion involves the frontal and subcortical regions, may show perseveration in cancellation tasks, repeatedly crossing out targets and adding irrelevant details and even complex drawings. Perseveration in patients with left neglect, however, occurs in the ipsilesional side of the sheet, namely, in the not (or less) neglected portion of space, where the preserved exploration takes place.^{5,6,30} These lateralized pathologic behaviors associated with left neglect clearly differ from the pattern observed in the current patients, particularly Patient 1, who not only do not show any evidence of omissions in drawing tasks but also add left-sided details (petals to the daisy), exhibiting, if anything, some sort of perseveration behavior in the contralesional side of space.

It could be also possible that the patients' behavior (drawing a larger left-hand side of objects, leftward overextension) reflects a compensatory disproportionate attention toward the contralesional side, during recovery from left spatial neglect. In the two patients, however, left neglect was mild and inconsistent across tasks. Furthermore, a compensatory behavior does not explain the underestimation of the size of left-sided objects.

The leftward contralesional expansion in drawing by copy and from memory bears some resemblance to the leftward biases in bisection performance, induced in patients with right neglect by optical illusions that displace the stimulus leftwards, such as the Judd figure with fins pointing rightward,³¹ or expand the left side of the stimulus, such as the Brentano figure with left-sided outward-projecting fins³² and the right-pointing triangle illusion³³. These data are consistent with the view that the pathologic mechanism bringing about the leftward expansion in drawing tasks found in the current two patients may involve a leftward enlargement or displacement. This, however, concerns the spatial medium, and not the perceived stimulus, as for visual illusion of length and displacement. In the drawing from memory task, where no physical stimulus is present, the patients' drawings are larger on the left side. In the

extension task, patients show a leftward overextension, toward an empty space, of a line presented to the right of the midsagittal plane. Furthermore, the hypothesis of a spatial medium pathologically expanded leftward predicts that left-sided stimuli are perceived as smaller than right-sided stimuli. Both patients underestimate the length of the left-sided rectangle in the perceptual matching task. By contrast, a leftward expansion of the stimulus, such as that brought about by the Brentano figure with a left-sided outward-projecting fin, makes its left side illusorily longer.³⁴ In right-brain-damaged patients with left neglect,³¹⁻³³ and particularly in patients without left half-visual field deficits,³⁵ the illusory leftward expansion or displacement of the stimulus reduces the rightward bias of neglect in line bisection. Conversely, a spatial medium pathologically expanded leftward would make left-sided stimuli, or the left part of a single stimulus such a line, shorter, increasing, if anything, the rightward bias in line bisection.³⁶ The very mild, and inconsistent across tasks, evidence of left spatial neglect found in the two patients suggests, however, that the leftward relaxation of the spatial medium and the rightward bias of left unilateral spatial neglect are largely independent pathologic phenomena.

In sum, the disproportionate enlargement of the left side of objects may be interpreted in terms of a horizontal anisometry of the medium for the representation of spatial relationships that was originally suggested to account for the paradoxical behavior of some right-brain-damaged patients with left neglect, who exhibit hyperextension when required to reproduce the perceived extent of a horizontal segment in a leftward direction.^{36,37} The hypothesis of a leftward relaxation of the medium predicts a left overextension in line extension tasks: The left side of the line would be perceived as shorter, being mapped onto a medium disproportionately relaxed leftward, and leftward movements would be then overextended.³⁶ Not all patients with left unilateral spatial neglect show contralesional hyperextension, however, and the suggestion has been made that left hemianopia may be a relevant factor.³⁸ In the current patients, by contrast, no visual field deficits were present, suggesting that contralesional hyperextension may be largely independent of both unilateral spatial neglect and visual field deficits. A leftward expansion of spatial representations may also bring about the pathologic leftward compensatory extension in drawing documented in this study: Left-sided objects (or the left half of symmetric objects) would be perceived as smaller compared with right-sided objects (or the right half of symmetric objects), being mapped onto a spatial medium expanded leftward. It may be finally noted that in the church drawn by Patient 1, the left-hand side of the windows, particularly the shutters, appear larger (figure 3B). Also, the left-hand side of the leaf located to the right of the daisy is greater than the right-hand side (figure 3A). These qualitative observations suggest that drawing may

be modulated by the intrinsic characteristics of the object.^{39,40}

The collateral finding that control subjects, both in copying and in drawing from memory tasks, draw the right-hand side of the daisy slightly larger may be seen, similar to the leftward error in line bisection, as a manifestation of pseudoneglect.⁴¹ The representation of the left-hand side of the daisy is larger in size, presumably owing to a physiologic leftward attentional bias, and this may result in a compensatory rightward expansion of the drawing.

Finally, the adding of left-sided petals, particularly by Patient 1, may be interpreted as perseveration or, more generally, as a case of productive behavior in a unilaterally disordered spatial medium. This would take place in the contralesional side of space, owing to the pathologic expansion of the medium in that direction, under relatively unconstrained conditions, such as drawing, that may release these productions. Studies in right-brain-damaged patients with ipsilesional perseveration associated with left spatial neglect^{5,6,42} suggest that damage to the frontal regions and anterior subcortical structures (caudate nucleus, putamen, frontal white matter) represent a possible neuropathologic counterpart to this productive, rather than defective, behavior, that may be conceived as an uncontrolled release of complex motor activities, such as drawing, in a disordered sector of space. The head of the caudate nucleus and the putamen are damaged in both patients, as well as the insular region. Right-brain-damaged patients with lesions including⁴² or restricted⁴³ to the insula may show productive impairments such as left somatoparaphrenia,^{42,43} ipsilesional perseveration, and other complex gratuitous productions in cancellation tasks.⁴² In sum, productive behaviors of right-brain-damaged patients in drawing tasks occur more frequently ipsilesionally^{5,6,30}, as contralesional left spatial neglect may be also present. However, perseveration may be found also contralesionally, as in the current patients, who show minimal evidence of neglect, inconsistent across tasks, and are able to fully explore the whole of extrapersonal space.

In 1905 Bonnier⁴⁴ reported that patients with peripheral vestibular disease may subjectively feel that their body, or parts of it, are disproportionately enlarged and interpreted this disorder as a pathologic expansion of the spatial representation of the body or of body parts (hyperschematia). This is probably the first report of a pathologic expansion of spatial representations, resulting in a disordered phenomenal experience of the size of the body or of body parts.⁴⁵ In right-brain-damaged Patients 1 and 2, the leftward expansion or relaxation concerns the medium for extrapersonal space, rather than the body. This disproportionately reduces the perceived size of contralesional objects, yielding a pathologic symptom complex that includes the disproportionate expansion of the left side of objects in drawing tasks, a leftward overextension in matching tasks with a left-

ward movement, and the underestimation of the lateral extension of left-sided objects in perceptual matching tasks. Perseveration behavior may be an additional component factor. In the two reported patients, the deficit, which may be termed contralateral hyperschemata for extrapersonal space, was associated with damage to the right hemisphere. This lateralization of the lesion, similar to the syndrome of spatial unilateral neglect,^{3,36} further supports a spatial nature of the disorder.

Acknowledgment

The authors thank Prof. Bernard Schott for comments on an earlier version of this manuscript, Dr. Jean Hubert Courjon for help with the microscope technique, and Mr. Jean-Louis Borach for technical support.

References

- Gainotti G. Constructional apraxia. In: Frederiks JAM, ed. *Clinical neuropsychology*. Amsterdam: Elsevier Science Publishers B.V., 1985: 491–506.
- Grossi D, Trojano L. Constructional and visuo-spatial disorders. In: Behrmann M, ed. *Handbook of neuropsychology*. 2nd ed. Amsterdam: Elsevier Science, 2001:99–120.
- Heilman KM, Watson RT, Valenstein E. Neglect and related disorders. In: Heilman KM, Valenstein E, eds. *Clinical neuropsychology*. 4th ed. New York: Oxford University Press, 2003:296–346.
- Vallar G. Spatial hemineglect in humans. *Trends Cogn Sci* 1998;2:87–97.
- Rusconi ML, Maravita A, Bottini G, Vallar G. Is the intact side really intact? Perseverative responses in patients with unilateral neglect: a productive manifestation. *Neuropsychologia* 2002;40:594–604.
- Na DL, Adair JC, Kang Y, Chung CS, Lee KH, Heilman KM. Motor perseverative behavior on a line cancellation task. *Neurology* 1999;52: 1569–1576.
- Critchley M. *The parietal lobes*. New York: Hafner, 1953.
- Guariglia C, Lippolis G, Pizzamiglio L. Somatosensory stimulation improves imagery disorders in neglect. *Cortex* 1998;34:233–241.
- Albert ML. A simple test of visual neglect. *Neurology* 1973;23:658–664.
- Wilson B, Cockburn J, Halligan PW. *Behavioural inattention test*. Titchfield, Hants: Thames Valley Test Co., 1987.
- Gainotti G, Messerli P, Tissot R. Qualitative analysis of unilateral spatial neglect in relation to laterality of cerebral lesions. *J Neurol Neurosurg Psychiatry* 1972;35:545–550.
- Castaigne P, Laplane D, Degos J-D. Trois cas de négligence motrice par lésion rétro-rolandique. *Rev Neurol* 1970;122:234–242.
- Diller L, Weinberg J. Hemi-Inattention in rehabilitation. The evolution of a rational remediation program. In: Weinstein EA, Friedland RP, eds. *Hemi-inattention and hemisphere specialization*. New York: Raven Press, 1977:62–82.
- Gauthier L, Dehaut F, Joanette Y. The Bells test: a quantitative and qualitative test for visual neglect. *Int J Clin Neuropsychol* 1989;11:49–54.
- Osterrieth PA. Le test de copie d'une figure complexe. *Arch Psychol* 1944;30:206–356.
- Rode G, Perenin MT. Temporary remission of representational hemineglect through vestibular stimulation. *Neuroreport* 1994;5:869–872.
- Schenkenberg T, Bradford DC, Ajax ET. Line bisection and unilateral visual neglect in patients with neurologic impairment. *Neurology* 1980; 30:509–517.
- Crawford JR, Garthwaite PH. Investigation of the single case in neuropsychology: confidence limits on the abnormality of test scores and test score differences. *Neuropsychologia* 2002;40:1196–1208.
- Crawford JR, Howell DC. Comparing an individual's test score against norms derived from small samples. *Clin Neuropsychol* 1998;12:482–486.
- Nico D. Detecting directional hypokinesia: the epidiascope technique. *Neuropsychologia* 1996;34:471–474.
- Coslett HB, Bowers D, Fitzpatrick E, Haws B, Heilman KM. Directional hypokinesia and hemispatial inattention in neglect. *Brain* 1990; 113:475–486.
- Milner AD, Harvey M, Roberts RC, Forster SV. Line bisection errors in visual neglect: misguided action or size distortion? *Neuropsychologia* 1993;31:39–49.
- Cohen L, Gray F, Meyrignac C, Dehaene S, Degos JD. Selective deficit of visual size perception: two cases of hemimicropsia. *J Neurol Neurosurg Psychiatry* 1994;57:73–78.
- Frassinetti F, Nichelli P, di Pellegrino G. Selective horizontal dysmetropia following prestriate lesion. *Brain* 1999;122:339–350.
- Kassubek J, Otte M, Wolter T, Greenlee MW, Mergner T, Luecking CH. Brain imaging in a patient with hemimicropsia. *Neuropsychologia* 1999;37:1327–1334.
- Azouvi P. Functional consequences and awareness of unilateral neglect: study of an evaluation scale. *Neuropsychol Rehabil* 1996;6:133–150.
- Halligan PW, Fink GR, Marshall JC, Vallar G. Spatial cognition: evidence from visual neglect. *Trends Cogn Sci* 2003;7:125–133.
- Marshall JC, Halligan PW. Within- and between-task dissociations in visuo-spatial neglect: a case study. *Cortex* 1995;31:367–376.
- Halligan PW, Marshall JC. Left visuo-spatial neglect: a meaningless entity? *Cortex* 1992;28:525–535.
- Manly T, Woldt K, Watson P, Warburton EF. Is motor perseveration in unilateral neglect "driven" by the presence of neglected left-sided stimuli? *Neuropsychologia* 2002;40:1794–1803.
- Ro T, Rafal RD. Perception of geometric illusions in hemispatial neglect. *Neuropsychologia* 1996;34:973–978.
- Vallar G, Daini R, Antonucci G. Processing of illusion of length in spatial hemineglect. A study of line bisection. *Neuropsychologia* 2000; 38:1087–1097.
- Shulman MB, Alexander MP, McGlinchey-Berroth R, Milberg W. Triangular backgrounds shift the bias of line bisection performance in hemispatial neglect. *J Neurol Neurosurg Psychiatry* 2002;72:68–72.
- Coren S, Giroux JS. Visual illusions. In: Held R, Leibowitz HW, Teuber H-L, eds. *Handbook of sensory physiology. Perception*. Heidelberg: Springer-Verlag, 1978:548–568.
- Daini R, Angelelli P, Antonucci G, Cappa SF, Vallar G. Exploring the syndrome of spatial unilateral neglect through an illusion of length. *Exp Brain Res* 2002;144:224–237.
- Bisiach E, Vallar G. Unilateral neglect in humans. In: Boller F, Grafman J, Rizzolatti G, eds. *Handbook of neuropsychology*. 2nd ed. Amsterdam: Elsevier Science B.V., 2000:459–502.
- Bisiach E, Rusconi ML, Peretti V, Vallar G. Challenging current accounts of unilateral neglect. *Neuropsychologia* 1994;32:1431–1434.
- Doricchi F, Angelelli P. Misrepresentation of horizontal space in left unilateral neglect. Role of hemianopia. *Neurology* 1999;52:1845–1852.
- Halligan PW, Marshall JC. When two is one: a case study of spatial parsing in visual neglect. *Perception* 1993;22:309–312.
- Mozer MC. Frames of reference in unilateral neglect and visual perception: a computational perspective. *Psychol Rev* 2002;109:156–185.
- Jewell G, McCourt ME. Pseudoneglect: a review and meta-analysis of performance factors in line bisection tasks. *Neuropsychologia* 2000;38: 93–110.
- Bottini G, Bisiach E, Sterzi R, Vallar G. Feeling touches in someone else's hand. *Neuroreport* 2002;13:249–252.
- Cereda C, Ghika J, Maeder P, Bogousslavsky J. Strokes restricted to the insular cortex. *Neurology* 2002;59:1950–1955.
- Bonnier P. L'aschématie. *Rev Neurol* 1905;13:605–609.
- Vallar G, Papagno C. Pierre Bonnier's (1905) cases of bodily "aschématie." In: Code C, Wallech C-W, Joanette Y, Lecours AR, eds. *Classic cases in neuropsychology, volume 2*. Hove, East Sussex: Psychology Press, 2003:147–170.

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Neurology 2006;67;1801-1808

DOI 10.1212/01.wnl.0000244432.91915.d0

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