

## REMISSION OF HEMINEGLECT AND ANOSOGNOSIA DURING VESTIBULAR STIMULATION

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**Abstract**—The effects of vestibular stimulation on extrapersonal and personal neglect and on awareness of disease were investigated in four patients with severe neglect and anosognosia. Neglect phenomena improved in all patients, while an effect on anosognosia was found in two cases. These preliminary findings indicate a possible role of vestibular stimulation on hemispheric activation.

### INTRODUCTION

THE FIRST case of transitory attenuation of neglect phenomena during vestibular stimulation was documented by SILBERPFENNIG in 1941 [11]. This finding has recently been confirmed by RUBENS [9], by testing unilateral neglect on tasks requiring visual exploration of extrapersonal space on a series of patients. These data are subject to two different interpretations: (a) it might be suggested that transitory reduction or elimination of visual neglect is due to ocular deviation towards the neglected field during ipsilesional vestibular excitation or contralesional vestibular inhibition; (b) the effect might be explained in terms of neurodynamic interactions involving brain structures subserving space representation.

To settle the question we started investigating the effect of vestibular stimulation on personal neglect phenomena, independently of visual control. Furthermore, in order to explore the consequences of vestibular stimulation on higher cognitive functions, we also decided to look for possible changes in awareness of disease, disorders of which are very closely related to hemineglect [2].

Given the obvious difficulties inherent in this program, we present a preliminary report, which it is hoped will stimulate further research along the same line.

### CASE REPORTS

*Case 1.* P.C., a 71 year old right-handed woman with 8 years of schooling and a long history of ischaemic heart disease, was admitted to hospital on 16 November, 1985 after the sudden onset of loss of strength in her left limbs. The patient was alert and cooperative, but easily distracted. A neurological examination revealed a left homonymous hemianopia on confrontation, left supranuclear facial palsy and left hypotonic hemiplegia with increased deep tendon reflexes. Touch and pain sensation was abolished on the left side. The patient did not respond to threatening gestures from the left and failed to notice people approaching or talking to her from the left side of the room. Head and gaze were both deviated to the right and did not cross the midline subsequent to verbal commands

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and visual pursuit. The patient denied the presence of any motor or visual impairment and stated she had come to hospital for a headache.

The CT scan performed at admission was negative. In the following days her condition deteriorated rapidly. She became unresponsive and died on 18 November, 1985. The pathologist reported a large cortico-subcortical softening involving the entire territory of the right middle cerebral artery. Unfortunately, the specimen was unsuitable for photographic reproduction.

*Case 2. A.D.*, a 69 year old right-handed hypertensive woman with 5 years of schooling, suddenly developed a loss of strength in her left limbs in the evening of 12 March, 1986. On admission to hospital the following day, the patient was alert and co-operative. A neurological examination revealed a left homonymous hemianopia, left supranuclear facial palsy and left hemiplegia. Touch and pain sensation was abolished on the left side and a steady right-sided deviation of the head and gaze, which never crossed the midline subsequent either to verbal command or visual pursuit, was also present. When addressed from the left side, the patient searched for the examiner turning her head and gaze further and further to the right. On acoustic double simultaneous stimulation (finger snapping) systematic extinction of the left stimulus was shown. Unilateral right-sided acoustic stimuli were correctly lateralized, while left-sided stimuli were displaced rightwards. The patient denied the presence of any motor or visual defect.

A CT scan performed on the day of admission revealed a large cortico-subcortical hypodense area involving the right temporoparietal region (Fig. 1). The insula, the lenticular nucleus and the internal capsule were involved; the right lateral ventricle was compressed.

*Case 3. G.S.*, a 57 year old right-handed hypertensive man with 8 years of schooling was admitted to hospital on 28 April, 1986 after the sudden onset of headache, slurred speech and loss of strength in his left limbs. On neurological examination the patient was alert, with a tendency to fall asleep when left alone. He had a mild parietal dysarthria without aphasic disturbances. On confrontation a left homonymous hemianopia was present. The patient suffered left hemiplegia with increased deep tendon reflexes and touch and pain sensation was abolished on the left side. Head and gaze were deviated rightwards, but given either verbal commands or pursuit movements the eyes could cross the midline, rapidly returning to the right side after stimulation. The patient denied any motor or visual problems, stating he was in hospital for "giddiness".

A CT scan on the day of admission revealed a right thalamocapsular haematoma (Fig. 2). Neglect recovered in about 1 week, but the hemiplegia remained unchanged and the patient was referred to a rehabilitation hospital 20 days after admission to our department.

*Case 4. S.C.*, a 48 year old right-handed hypertensive man with 8 years of schooling, was admitted to hospital on 10 June, 1986, after the sudden onset of loss of strength in his left limbs. The patient was alert and co-operative, but tended to fall asleep when left unstimulated. A neurological examination revealed a left homonymous hemianopia on confrontation, supranuclear left facial palsy and left hypotonic hemiplegia with increased deep tendon reflexes. Anaesthesia to touch and pain on the left side completed the clinical picture. Head and gaze were deviated to the right, but the patient could temporarily move the eyes towards the left on both verbal command and visual pursuit. The patient denied any motor or visual defect and stated he had come to hospital for treatment for a headache and because his wife had noticed that his mouth was distorted.

A CT scan performed on the day of admission showed a hazy hypodensity in the right insular region, and, when repeated after 7 days, a large cortico-subcortical hypodense area involving the whole territory of the right middle cerebral artery was revealed (Fig. 3). The insula, the lenticular nucleus, the internal capsule and the thalamus were involved. The clinical course was characterized by two episodes of severe deterioration of consciousness, which recovered after intravenous mannitol. Neglect and anosodiaphoria were still present 20 days after admission.

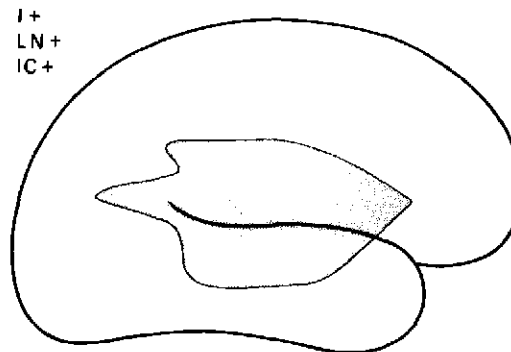


FIG. 1. Lateral map [8] of the ischaemic lesion of case 2. I+, LN+, IC+: insula, lenticular nucleus and internal capsule involved.

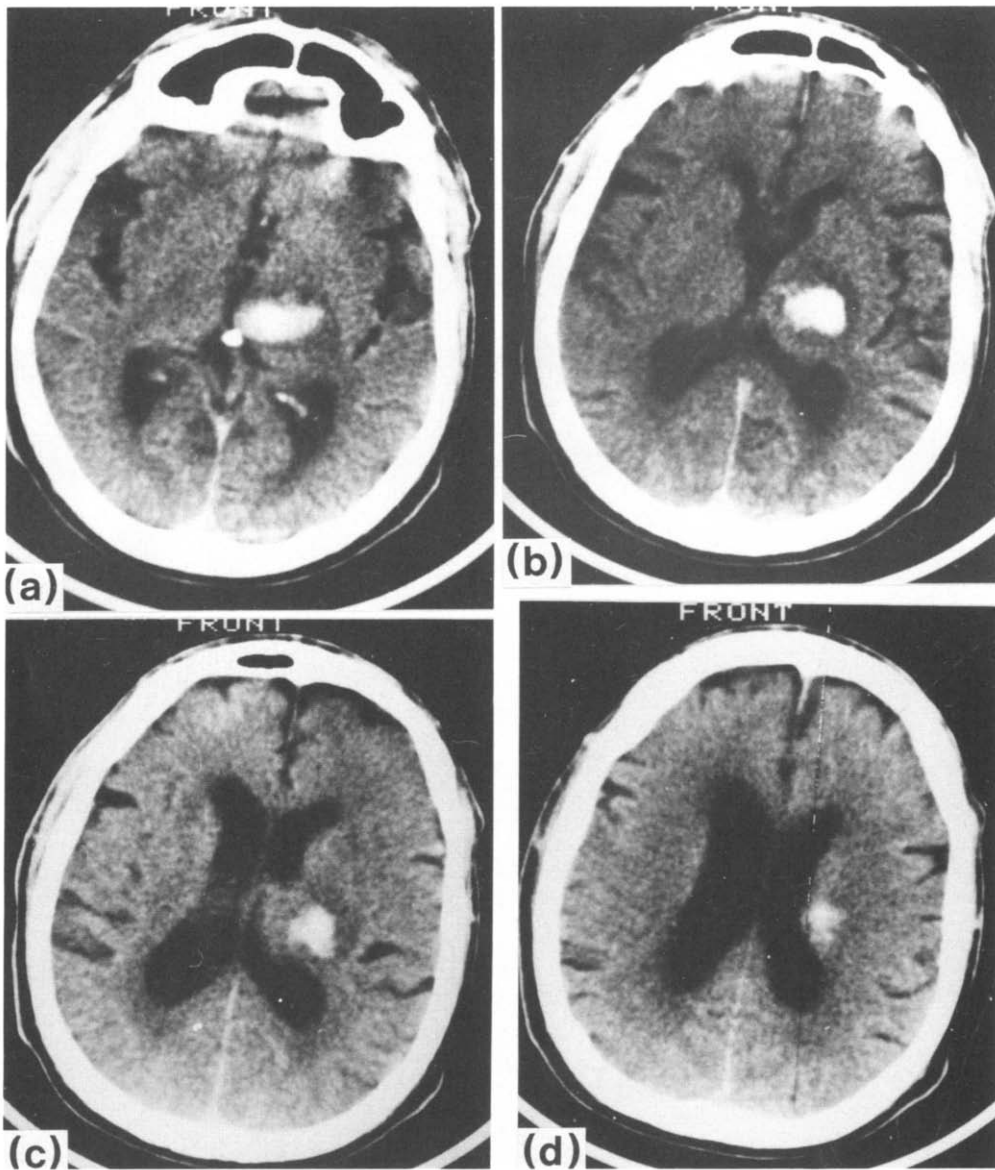


FIG. 2. CT Scan of case 3. Haematoma, surrounded by perifocal oedema, involving the right thalamus, the genu and the posterior limb of the internal capsule and the periventricular subcortical white matter.



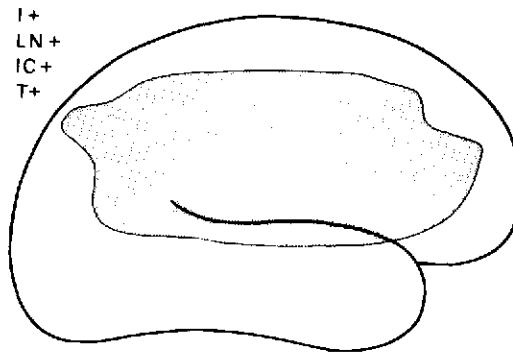


FIG. 3. Lateral map [8] of the ischaemic lesion of case 4. I+, LN+, IC+, T+: insula, lenticular nucleus, internal capsule and thalamus involved.

## METHOD

The following procedure was used to examine all four patients, within two days after the onset of the disease.

### Baseline assessment

1. *Extraperpersonal neglect* was evaluated by a circle crossing test [3] in cases 2, 3 and 4 and by a line crossing test [1] in case 1. The patients crossed out the items printed on a sheet of paper using their right unaffected hand. The number of omitted circles or lines was recorded.

2. *Personal neglect*. The examiner asked the patient, who was lying on his/her back, with the arms extended along the sides of the trunk, to touch his/her left hand with the right. A four-point scale was used [6]: 0 (target promptly reached), 1 (target reached with hesitation and search), 2 (search interrupted before the target is reached), 3 (no movement towards the target). Personal neglect was assessed with eyes open in case 1, with eyes both shut and open in cases 2, 3 and 4. In cases 2 and 4 the eyes shut condition was examined first.

3. *Anosognosia* for hemiplegia (upper and lower limbs) and for hemianopia was rated by a four-point scale [6]: 0 (disorder spontaneously reported or mentioned after a general question about the patient's complaints), 1 (disorder reported only following a specific question about the patient's deficit), 2 (disorder acknowledged only after having been demonstrated through routine techniques of neurological examination), 3 (disorder not acknowledged).

### Immediate post-stimulation assessment

The same procedures were repeated immediately after *caloric stimulation*. In cases 1, 2 and 4 the left external ear canal was irrigated with 20 cc of iced water for 1 min. In case 3 the right ear was irrigated with 20 cc of warm water, due to the presence of a great amount of cerumen in the left ear. In all patients caloric stimulation evoked a brisk nystagmus to the right. Irrigation seemed to produce some discomfort in all four patients, who however were unable to explain the nature of their unpleasant sensations and did not report vertigo or sense of displacement.

### Delayed post-stimulation assessment

The same procedures were repeated about 15 min after caloric stimulation.

## RESULTS

The results are summarized in Table 1. In the baseline condition, all four cases showed very severe *extraperpersonal neglect*, being able to cross only the more extreme right-sided items. All four patients showed a temporary reduction of neglect after caloric stimulation, as demonstrated by an increase in the number of circles or lines crossed out, followed by return to the base level after 15 min.

*Personal neglect* improved in all four patients. Case 1, who did not perform any movement towards the left hand in the basal condition, reached the target after stimulation, even though

Table 1. Effects of caloric stimulation on extrapersonal neglect, personal neglect and anosognosia in four right hemisphere damaged patients.

Case	1			2			3			4		
	I	II	III	I	II	III	I	II	III	I	II	III
Assessment												
Extrapersonal neglect % omissions	80	53	76	76	38	76	69	23	61	84	61	84
Personal neglect												
eyes closed	—	—	—	1	0	1	2	0	2	2	1	1
eyes open	3	1	1	1	0	1	2	0	2	1	0	0
Anosognosia												
upper limb	3	3	3	3	3	3	3	1	1	3	2	1
lower limb	3	3	3	3	3	3	3	3	3	3	2	1
visual field	3	3	3	3	3	3	3	3	3	3	3	3

I, baseline assessment; II, immediate post-stimulation assessment; III, 15 min delayed post-stimulation assessment.

with hesitation and search. The improvement was still present after 15 min. The task was performed only in the eyes open condition. This patient, who before stimulation and in the delayed post-stimulation assessment, accepted the examiner's arm as her own, could recognize it correctly immediately after stimulation. Case 2, who reached the left hand with some search in the basal condition, was fully accurate after stimulation. Case 3 attempted to reach the left hand without success in the baseline condition, but was fully accurate after stimulation. In neither patient was there any difference between the eyes open and eyes shut conditions and personal neglect was again present in the delayed post-stimulation assessment. Case 4 failed to touch his left hand in the eyes shut condition, but after some searching was successful in the eyes open condition. His performance improved after caloric stimulation, and this improvement continued into the days following the test.

All four patients were severely *anosognosic* for both motor and visual deficits. In cases 1 and 2 anosognosia was not affected by caloric stimulation. On the other hand, anosognosia for hemiplegia was reduced after stimulation in cases 3 and 4. Case 3, during the immediate post-stimulation assessment, only after specific inquiry admitted that the left hand was weaker than the right. He persisted in denying any deficit in the lower limb after specific questions, and an invitation to raise it from the bed and a demonstration of the disorder. Case 4 acknowledged the motor deficits immediately after stimulation only when asked to raise his left limbs from the bed. After 15 min, questioned specifically about his left limbs, he said: "I don't know why, I have always been able to move them, but now they are blocked: it is as if my brain is no longer able to command them". This behavioral change was still present in the following days.

## DISCUSSION

Our first results confirm the observations made by SILBERPFENNIG [11] and RUBENS [9] relative to visual neglect.

Remission of personal neglect was found in all patients. This may suggest that even in the eyes open condition the effect was not simply due to an enlargement of the gaze field during vestibular stimulation. In fact, the action required for the assessment of personal neglect is usually executed without apparent visual control [5]; moreover, unlike extrapersonal

neglect which soon reappears once the effect of vestibular stimulation is over, personal neglect may show a more enduring improvement. However, the reappearance of personal neglect in the post-stimulation assessment of patients 2 and 3 (Table 1), shows that remission of the symptom during stimulation was not due to repeated testing. An oculomotor explanation of the lessening of the level of personal neglect during vestibular stimulation, on the other hand, is even more convincingly contradicted by the attenuation of the disorder in the eyes shut condition: this is especially true for patients 2 and 4, in which this condition was tested first.

A vestibular stimulation effect on anosognosia was found in two patients only. A dissociation between neglect and anosognosia has already been found by BISIACH *et al.* [6] and, as argued elsewhere [2], it does not undermine a unitary conception of the syndrome. In the two positive instances, the remission of anosognosia outlasted the period of vestibular stimulation. It would however be premature to venture any interpretation of this finding, as well as of its relation with the parallel trend regarding personal neglect.

Whoever is acquainted with anosognosia, and knows how obdurate it may be, cannot fail to be impressed by the fact that a pathological belief which is impervious to any rational counterargument [4] and indeed in our patients resisted a straightforward demonstration of the severe neurological disorder, actually yielded to vestibular stimulation in patients 3 and 4. Since, for practical reasons, a control condition was not included, an interpretation of the reduction of anosognosia in terms of repeated questioning cannot be ruled out altogether. Such an interpretation, however, is most implausible in view of the firm resistance to any kind of persuasion of the pathological beliefs which characterize severe anosognosia. Our findings, if confirmed, should eradicate any inclination to interpret anosognosia in terms of "general confusion" or "intellectual deterioration", as well as more elaborate accounts offered by authors of psychodynamic persuasion (see [4, 6] for further arguments against these interpretations).

At present, it would be untimely to speculate through which neural mechanism vestibular stimulation improves hemineglect, since we do not even know whether other kinds of stimulation may produce similar effects. The recent regional cerebral blood flow study by FRIBERG *et al.* [7] has shown a consistent focus of cortical activation contralateral to vestibular stimulation, located in the posterior-superior region of the temporal lobe, i.e. in close proximity to an area which is known to be strictly related to egocentric space representation [12]. Due to technical constraints, the authors were not able to measure regional blood flow from the corresponding region ipsilateral to the stimulated ear. Two possible mechanisms by which vestibular stimulation could influence neglect phenomena in right brain damaged patients are ipsilateral cortical activation and activation of the intact contralateral hemisphere. However, even if ipsilateral activation were proven to occur in the normal brain, any possible effect on the areas related to spatial representation in the right hemisphere appears unlikely in patients with extensive damage to these regions (cases 1, 2, 4). In case 3 the cortex was unaffected on CT assessment. There is however evidence that a major cortical dysfunction is present in neglect patients with subcortical stroke lesions [10]. Thus it appears more likely that the (more or less temporary) regression of these symptoms which was obtained in our patients by creating an imbalance of the two vestibular systems was due to activation of the hemisphere opposite to the relatively hyperactive vestibular system, i.e. the left, unlesioned hemisphere. This might support the hypothesis that structures of the left hemisphere tend to compensate for the disorder in case of extensive, irreversible right hemisphere damage.

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