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SHORT COMMUNICATIONS

Signs of spatial neglect in unilateral peripheral vestibulopathy

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Abstract

Background and purpose: In this study, the question of whether egocentric representation of space is impaired in chronic unilateral vestibulopathies was examined. The objective was to test current theories attributing a predominant role to vestibular afferents in spatial cognition and to assess whether representational neglect signs are common in peripheral vestibular loss.

Methods: The subjective straight-ahead (SSA) direction was investigated using a horizontal rod allowing the translation and rotation components of the body midline representation to be dissociated in 21 patients with unilateral vestibular loss (right, 13; left, eight) and in 12 healthy controls.

Results: Compared to the controls, the patients with unilateral vestibulopathy showed a translation bias of their SSA, without rotation bias. The translation bias was not lateralized towards the lesioned side as typically found for biases reported after unilateral vestibular loss. Rather, the SSA bias was rightward whatever the side of the vestibular loss. The translation bias correlated with the vestibular loss, as measured by caloric response and vestibulo-ocular reflex gain, but not with the subjective visual vertical or the residual spontaneous nystagmus.

Conclusion: The present data suggest that the dysfunctions of neural networks involved in egocentred and allocentred representations of space are differentially compensated for in unilateral vestibular defective patients. In particular, they suggest that asymmetrical vestibular inputs to cortical regions lead to representational spatial disturbances as does defective cortical processing of vestibular inputs in spatial neglect after right hemisphere stroke. They also highlight the predominant role of symmetrical and unaltered vestibular inputs in spatial cognition.

KEYWORDS
spatial neglect, subjective straight-ahead, unilateral vestibular loss

BACKGROUND

Space perception is based on the integration of signals from vestibular, visual and somatosensory systems [1,2] As a result of this integration, individuals become aware of the displacements and positions of their body and body parts as well as of the locations of objects in space. Brain lesions can produce severe deficits in the representation of personal or extra-personal space as demonstrated by the syndrome of unilateral (left) spatial neglect after (right) hemisphere stroke [3] Clinically, neglect entails a variety of deficits in spatial cognition (e.g., [4]). It may produce both egocentric deficits (affecting left space in a body-centred reference frame) and allocentric deficits (affecting left space in an object-centred reference frame). Different deficits may reflect sensory perceptual vs. motor exploratory aspects. Manifestations of egocentric neglect can be

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both motor exploratory and sensory perceptual whereas allocentric signs are more generally perceptual [5,6] Vestibular stimulation was shown to modulate spatial neglect [7] Moreover, symptoms similar to those of neglect were observed in patients with dysfunctions of the peripheral or central components of the vestibular pathways, which lead to the hypothesis that vestibular dysfunction might contribute to spatial biases in neglect [8]

Thus, it was repeatedly reported that neglect patients had an abnormal posture, tilted or turned towards the lesioned side, and that their verticality perception was biased [6] In the same vein, the literature on vestibular compensation classically states that unilateral peripheral lesions induce a tonic imbalance in vestibular inputs resulting in postural deviations or abnormal verticality perception [1,8]

Concerning body representation, neglect patients showed a compression, with the left hemibody perceived as 'narrower' than the right, as well as a shift of the subjective body midline towards the lesioned side [5,6,9] In unilateral vestibular defective patients, the issue was less investigated. Nevertheless, a recent study described a distorted representation of the extra-personal space in the acute stage after right vestibular loss including a shift towards the lesioned side and a compression towards the contralesioned hemifield [10] Moreover, the study by Saj et al. [11] suggested that the subjective body midline is shifted at the chronic stage after a total unilateral vestibular neurotomy.

Apart from these extreme cases of total unilateral vestibular loss, whether the body representation is commonly impaired in unilateral vestibulopathies has been ignored. This representation was thus assessed using a variant of the subjective straight-ahead (SSA) test [5,6,9] in which the participants are required to put a horizontal luminous rod on their subjective midsagittal plane [5,6,11]

MATERIALS AND METHODS

Participants

Twenty-one patients with a strictly unilateral vestibular loss (VL) participated in the experiment. All of them were tested in the chronic stage of their disease. Thirteen had a left VL (LVL) (mean age 57.2 ± 11.4; education level 12.4 ± 3.1; onset of the disease 3.3 ± 3.2 years; 13 right-handed; 11 right-dominant eye) and eight a right VL (RVL) (mean age 51.0 ± 17.9; education level 12.1 ± 3.8; onset of the disease 2.4 ± 1.8 years; eight right-handed; six right-dominant eye). The two patient groups were similar for education level (RVL vs. LVL, $p = 0.43$). Seventeen were enrolled with a vestibular schwannoma (Koos classification grade from 1 to 3) and four patients with Menière's disease. These two types of patients do not differ in terms of their clinical status (Table 1). Patients with additional motor, cognitive or oculomotor deficit were not included in the study. None was under antivertigo medication. All patients underwent otoneurological examinations including bithermal caloric tests, a vestibulo-ocular test on a rotating chair (leading to a vestibulo-ocular reflex [VOR] gain for both ears), spontaneous nystagmus measures in darkness and subjective visual vertical (SVV) (Table 1). The SVV was recorded in a sitting condition in order to allow a direct comparison with the SSA recordings. The patients with left and right vestibular loss did not differ in terms of their clinical status (Table 1). They were compared with 12 healthy control (C) subjects (mean age 51.7 ± 14.5; education level 17.0 ± 3.9; 12 right-handed; eight right-dominant eye) of similar age (C vs. LVL, $p = 0.15$; C vs. RVL, $p = 0.46$; RVL vs. LVL, $p = 0.17$), but education level differed from the control group ($p \le 0.05$). All participants were either emmetrope or had corrected vision. They signed an informed consent form, approved by the Ethical Committee of the North Hospital of Marseille, following the principles outlined in the Helsinki Declaration.

Apparatus

Participants sat upright, facing a horizontal metal rod (Figure 1), centred 50 cm in front of them, which could be simultaneously rotated in the yaw plane and translated along a 100-cm-wide slit in a horizontal plate located at navel height. The head was held up in the trunk direction. A disc mounted between the rod and the plate avoided haptic cues. Two potentiometers gave the rotation angle (degree, positive value for clockwise orientation) and the translation (mm, positive value for rightward displacement). Ten red light emitting diodes inserted in its upper side made the rod visible in darkness. The whole apparatus was centred relative to the body midline.

Procedure

The participants were instructed to imagine a line starting from the navel and extending away straight ahead of the trunk, and to place the rod on this virtual line. The adjustments were carried out in darkness, the rod being handled at its centre with the right hand. Before each trial, the rod was initially translated to −15 or +15 cm and rotated to −45° or +45°. The order of the four trials (one for each initial position) varied across subjects.

Statistical analysis

Separate analyses of variance were performed on SSA translation and rotation, including the between-factor 'group' (LVL, RVL, C). Pearson's coefficient was used to assess the relation between clinical parameters and SSA performance. The significance threshold was $p \le 0.05$.

RESULTS

Figure 1 shows the individual performance of patients in both translation and rotation with respect to the midsagittal plane. The translation significantly differed between the groups ($F(2, 30) = 7.80$,

TABLE 1 Demographic and clinical data of patients

Patient	Group	Gender	Age (years)	Delay since onset (years)	Aetiology	Koos classification grade	Hearing loss (dB)	Vestibular deficit (%)	VOR gain	Spontaneous nystagmus		SVV (degrees)
										Horizontal (°/s)	Vertical (°/s)	
L01	LVL	F	66	2	VS	2	46	88	0.39	2.1	0.2	1.0
L02	LVL	F	59	5	VS	3	45	100	0.56	0.1	0.1	2.2
L03	LVL	M	72	3	VS	3	65	100	0.21	0.4	0.8	1.1
L04	LVL	M	40	3	MD	n/a	25	41	0.48	0	0	−3.3
L05	LVL	F	29	1	VS	1	88	25	0.53	0.3	0	1.3
L06	LVL	F	65	1	VS	3	48	100	0.62	2	2.3	−0.4
L07	LVL	F	57	1	VS	3	49	82	0.37	0.8	0.5	3.0
L08	LVL	M	56	13	MD	n/a	41	31	0.41	0	0	0.0
L09	LVL	M	55	3	VS	3	43	81	0.28	0.8	0.1	0.4
L10	LVL	M	55	3	VS	2	24	100	–	–	–	0.0
L11	LVL	F	64	5	VS	3	26	100	0.48	0	0.7	−0.4
L12	LVL	F	61	1	VS	2	75	100	0.59	0	0	−3.2
L13	LVL	F	64	2	VS	2	36	23	0.56	0.1	0	0.1
R01	RVL	F	68	1	VS	2	20	76	0.63	1.9	1.4	0.8
R02	RVL	M	52	2	VS	2	63	54	0.12	0.1	0	1.4
R03	RVL	M	48	2	VS	3	21	40	0.45	0.3	0.1	−0.4
R04	RVL	F	58	6	MD	n/a	59	58	0.48	0.3	0.1	1.8
R05	RVL	F	26	2	VS	2	21	59	0.38	0.3	1.3	3.0
R06	RVL	M	23	1	VS	2	58	63	0.44	0.3	0.7	−0.2
R07	RVL	M	66	1	VS	3	64	42	0.55	0.2	0.5	−3.3
R08	RVL	M	67	4	MD	n/a	74	100	0.28	0.6	0.1	4.7

Abbreviations: F, female; LVL, left vestibular loss; M, male; MD, Menière's disease; n/a, not applicable; RVL, right vestibular loss; SVV, subjective visual vertical (negative value, anticlockwise deviation); VOR, vestibulo-ocular reflex; VS, vestibular schwannoma.

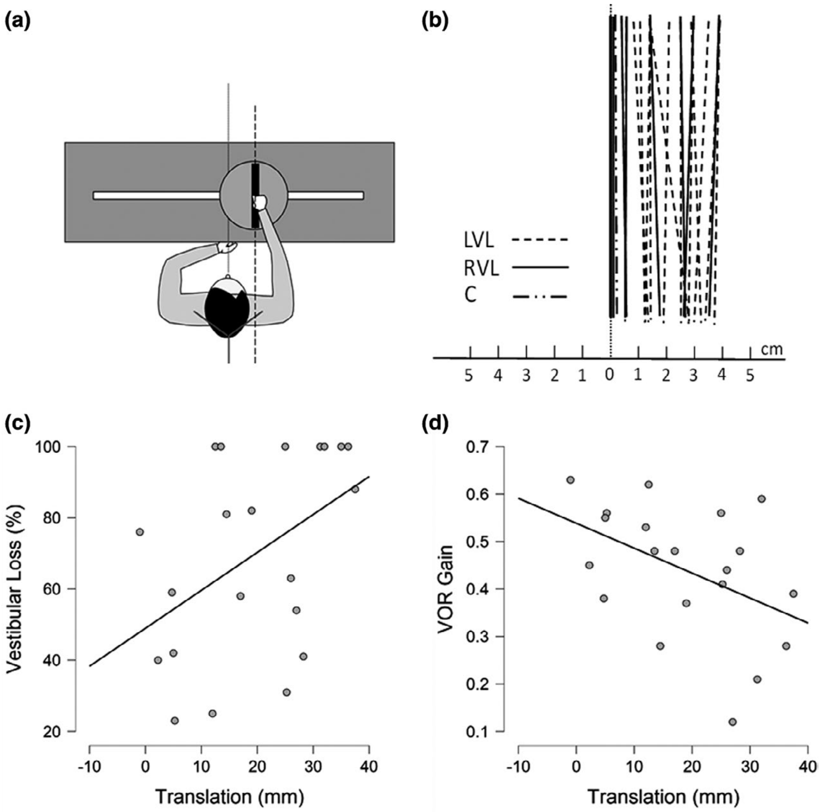

FIGURE 1 (a) Participants, sitting in darkness, had to place a luminous rod in their midsagittal plane. The rod could be simultaneously rotated and translated. The task was to indicate the direction straight ahead of their navel. (b) Subjective straight-ahead direction indicated by each patient. Full lines, right vestibular loss (RVL) patients; dashed lines, left vestibular loss (LVL) patients; dotted and dashed line, mean of control group (C). (c), (d) Relation between translation deviation and (c) vestibular loss and (d) VOR gain

$p = 0.002$). The contrast between the C group (mean ± confidence interval [CI] 3.4 ± 7.2 mm) and the patients (19.4 ± 5.2 mm) was significant ($F(1, 30) = 11.79$; $p = 0.002$), whilst the contrast between LVL (22.4 ± 5.7 mm) and RVL (11.6 ± 8.8 mm) patients was not ($F(1, 30) = 2.03$; $p = 0.164$). In 19 patients out of 21, the rod was translated more rightward than the mean position of controls.

The SSA translation correlated with the vestibular loss as evidenced by the caloric test (the greater the VL, the greater the translation, $r(19) = 0.46$; $p = 0.037$) and the VOR gain (the lower the VOR gain, the greater the translation, $r(18) = -0.45$; $p = 0.045$). No correlation was found between SSA translation and SVV, horizontal spontaneous nystagmus, hearing loss, delay since onset, or age.

Regarding the rotation of the SSA, the mean errors were weak and did not differ between the patients (0.14° ± 1.04°) and the controls (0.36° ± 0.76°) ($F(1, 31) = 0.42$; $p = 0.521$).

DISCUSSION

The original finding of this study consisted in a translation of the SSA of patients suffering from chronic unilateral vestibular deficits. This SSA bias, which was rightward whatever the lesioned side, contrasted with the lack of any bias of the SVV. The size of the bias appeared to correlate with indicators of deficit intensity as provided by the caloric response or the VOR gain.

The SSA translation bias appeared here rather systematic, as was also the case in patients who neglected left space after a right hemisphere stroke [5,6] As in neglect patients, no systematic rotation bias occurred here. Moreover, the SSA deviation was associated neither to the change of gaze (slow phase of nystagmus) nor to the hearing loss. It was found here that, at the difference with allocentred error (SVV) mostly present in acute stages of vestibular diseases, the egocentred error (SSA translation) did not seem to be subject to compensatory mechanisms. Moreover, the SSA test appeared effective to detect dysfunctions in body spatial representation of vestibular patients since 48% of the patients had rightward deviation beyond the range of controls, in line with a recent study using paper-and-pencil neglect tests that obtained a positive outcome in 32% of the patients with unilateral peripheral vestibulopathy [12]

The rightward deviation of the SSA is in line with the outcome of a previous paper [12] which described hemineglect symptoms in unilateral peripheral vestibulopathy using the bisection task. However, those authors found no difference in the frequency of hemispatial neglect between patients with right and left unilateral peripheral vestibulopathy and no definite lateral asymmetry in neglect manifestation. Here, in contrast, a rightward deviation independent of the side of the unilateral peripheral vestibulopathy was found. These differences could be related to the fact that the patients included by those authors suffered from an acute vestibular neuritis, whilst those of the present study were tested at a chronic stage of their disease. Nevertheless, beyond these differences, the fact that both studies showed neglect signs in vestibular patients could be related to the similarity of the SSA and bisection tasks. Indeed, it was shown in a previous study that the performances in these representational tasks strongly correlated in neglect patients and it was claimed that both consist in searching the virtual limit separating the two halves of a space [13] Furthermore, in another study where attentional cancellation tasks were privileged, no neglect signs were found in vestibular patients [14] Taken as a whole, these data suggest that the representational and attentional tasks assess different components of spatial cognition.

Our results argue in favour of signs of spatial neglect in unilateral peripheral vestibulopathy related to changes in vestibular inputs through the vestibulocortical pathways. Interestingly, the consequences of peripheral vestibular tone imbalance do not drastically differ from those of cortical imbalance for body orientation representation. Taken as a whole, the data at hand suggest that abnormal cortical processing of vestibular inputs after right hemisphere stroke might contribute to spatial neglect and also that asymmetrical abnormal vestibular inputs to cortical regions may lead to subjective spatial disturbances. The fronto-parietal components of the right neural networks lesioned in neglect were shown to involve the parietal lobe, the middle temporal gyrus and the superior frontal gyrus when egocentric coding deficits were present, and the insula, the inferior parietal gyrus and the thalamus when balance deficits occurred [6] All of these areas are known to be highly innervated by afferents originating in the vestibular system [15] All in all, the present data are in line both with the notion of a predominant role of vestibular afferents in spatial cognition [16] and could rather concern the representational component, and with the vestibular hypothesis of neglect.

In conclusion, these findings provide strong evidence that the vestibular system is involved in the representation of the body orientation in space.

CONFLICT OF INTEREST

The authors report no conflicts of interest.

DISCLOSURE

None.

DATA AVAILABILITY STATEMENT

Data are available on reasonable request.

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