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Diplopia from Skew Deviation in Unilateral Peripheral Vestibular Lesions

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Vertical diplopia from skew deviation is well described in brainstem lesions. The phenomenon can also result from peripheral vestibular lesions. During the past years, we have observed these ocular changes in the acute stage following unilateral vestibular neurectomy and labyrinthectomy ($n = 13$), as well as in a series of patients suffering from idiopathic sudden unilateral peripheral vestibular or cochleo-vestibular deficit ($n = 5$). Diplopia from skew deviation was noted immediately following ablative vestibular procedures; in patients with idiopathic vestibular deficit, it was observed as an associated sign. In all patients, clinical evaluation revealed an acute unilateral peripheral vestibular loss, with spontaneous nystagmus toward the unaffected ear and absence of nystagmic response to caloric testing on the affected ear. Skew deviation was mesured using the Hess-Weiss test, which is based on the haploscopic principle. Static visual vertical was evaluated with the original methods of vertical frame and Maddox rod techniques. Photographs were made of the ocular fundi, to measure the degree of cyclotorsion of both eyes. In our patients, we found skew deviation with hypotropia of the eye that was ipsilateral to the affected ear and conjugated cyclotorsion and tilt of the static visual vertical on the side of the affected ear. Skew deviation was the first sign to disappear within a few days; conjugated cyclotorsion and tilt of the static visual vertical persisted for weeks to months. The eye-head postural reaction, consisting of head tilt, conjugated eye cyclotorsion, skew deviation, and alteration of vertical perception directed toward the side of the lesion, is known as the Ocular Tilt Reaction (OTR). The mechanism is presumably related to a lesion of the otolithic organs and/or to changes in the afferent graviceptive pathways. In man, the OTR is often mild and unrecognized, masked by spontaneous nystagmus and marked neuro-vegetative symptoms. Our observations indicate that skew deviation, as a part of the OTR, occurs in patients with sudden peripheral vestibular lesions, whether surgical or non-surgical in origin. *Key words: vertigo, vertical diplopia, skew deviation, ocular tilt reaction, peripheral cochleo-vestibular loss, vestibular neuritis, static visual vertical, otolithic function.*

INTRODUCTION

In human subjects, vertical diplopia from skew deviation is well known in several types of brainstem lesion (1–4). During the past few years, however, this phenomenon has also been observed in an acute stage following unilateral peripheral vestibular surgery, such as labyrinthectomy or vestibular neurectomy (5–9) and in an isolated case of stapedectomy (10). In such cases, skew deviation was considered to be part of the eye-head postural reaction known as the ocular tilt reaction (OTR).

In the literature on non-surgical peripheral vestibular dysfunction, only isolated reports of skew deviation have been reported (11, 12). We have observed the phenomenon in several patients following sudden unilateral peripheral vestibular loss (13, 14), so-called vestibular neuritis (15).

In this paper, we report our findings in a series of patients who developed skew deviation as well as the other manifestations of OTR following either a surgical or non-surgical unilateral peripheral vestibular lesion. Skew deviation, abnomalities of the static visual vertical (SVV), conjugated cyclotorsion and head tilt were measured with a battery of specially designed oto-neuro-ophthalmological tests.

MATERIAL AND METHODS

Vertical diplopia resulting from skew deviation was observed in 13 patients suffering from Meniere's disease (5 men and 8 women, aged from 21 to 64 years) who were operated on by vestibular neurectomy ($n = 8$) or labyrinthectomy ($n = 5$) between 1993 and 1995 at the University Clinic of ENT, Head and Neck Surgery, in Berne. Skew deviation was also observed in 5 patients (3 men and 2 women, aged from 27 to 71 years) suffering from vestibular neuritis in 1992 at the Department of Clinical Neurosciences of the Geneva University Hospital (13) as well as in a 59-year-old woman with a sudden unilateral peripheral cochleo-vestibular loss, evaluated in Berne in 1993 (14). In all these patients, skew deviation and the other OTR manifestations were measured, and the results were compared with data from 32 normal subjects (20 men and 12 women, aged from 7 to 50 years) used as controls.

Skew deviation, and the different manifestations of OTR, were measured using the following specially designed oto-neuro-ophthalmological tests:

Measurement of skew deviation with the Hess-Weiss test. This test evaluates deviation of the visual axes based on the haploscopic principle (13). The patient

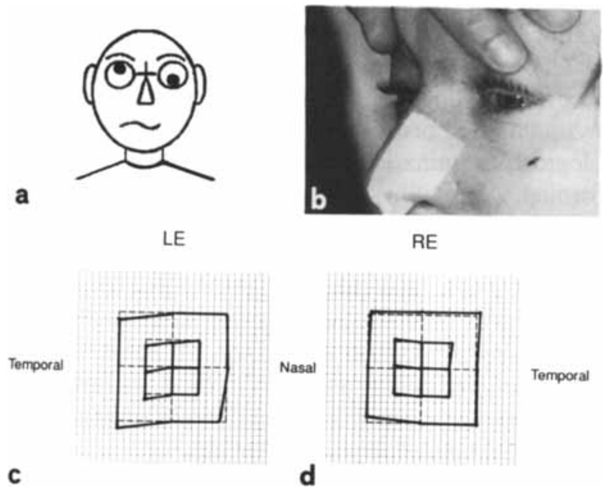

Fig. 1. Evaluation of skew deviation with the Hess-Weiss test.Vertical strabismus, or skew deviation, consists of hypotropia in the eye ipsilateral to the operated ear (*a*). Left hypotropia in a patient one hour after left vestibular neurectomy (*b*). At day 2nd after left vestibular neurectomy, the Hess-Weiss test (*c*,*d*) showed a slight hypotropia in the left eye, more pronounced in gaze left (*c*). At day 5th post-operatively, the vertical deviation wass markedly reduced.

is presented with a square white screen, marked with a series of red lines defining squares subtending approximately 5 diopters when viewed at a distance of 50 cm. A red dot is located at the zero point of this coordinate system and also at each intersection of the 20- and 40-diopter lines with one another and with the corresponding vertical and horizontal lines. These locations form an inner and an outer square. The

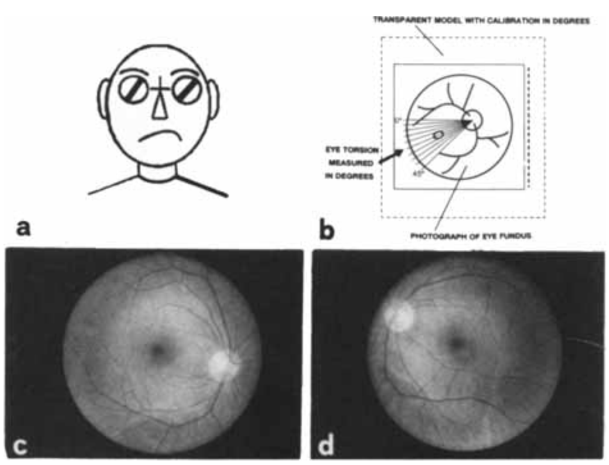

Fig. 2. Measurement of static visual vertical using the binocular vertical frame technique (*a*) and the monocular technique using the modified Maddox rod (*b*) with a dental bite. To ensure that the frame was horizontal, a spirit level was added to the transverse bar of the Maddδx rod, as well as to the dental bite.

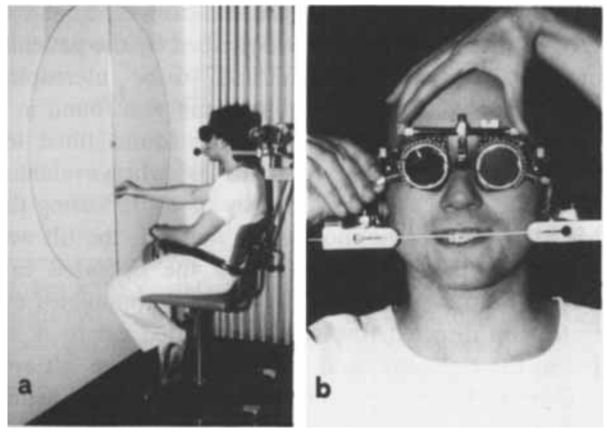

Fig. 3. Measurement of conjugated cyclotorsion of the eyes with ocular fundus photographs. The degree of cyclotorsion is quantified by comparing the papillo-macular axis tilt with the horizontal, using a graduated ruler (*b*). Ocular fundus photographs in patient Nr. 7 (Table I), one week after left vestibular neurectomy showed a leftward conjugated cyclotorsion, with 10° incyclotorsion in the right eye (*c*) and 20° excyclotorsion in the left eye (*d*).

patient is seated 50 cm from the screen with his head fixed on a headrest. He wears red-green goggles, and is provided with a red light pointer. The squares are seen with the eye fitted with the green filter, and the red light spot with the other eye. The positions indicated by the patient are plotted on a small chart showing a reduced copy of the screen, and are then connected by straight lines. The patient is first tested using the red filter in front of the right eye, and results are marked in the right eye grid. To change fixation the goggles are reversed, the red filter being placed in front of the left eye. The results are then plotted in the left eye grid (Fig. 1).

Measurement of static visual vertical with a vertical frame (binocular method) and a modified Maddox rod (monocular method). Using the vertical frame method the patient is seated in front of a large white vertical screen with a central pivoting ruler. A dental bite is used to ensure an upright head position, as shown in Fig. 2. The patient's visual field is restricted to the center of the board by an appropriate frame. He is asked to adjust the ruler along the axis of his subjective vertical perception. The ruler's tilt, in degrees, is measured against a graduated scale on the edge of the board.

The modified Maddox rod technique is based on the use of Maddox glasses, which transform the image of a luminous point into an illuminated line which is perpendicular to the axis of the Maddox rod. The patient is asked to adjust the illuminated line along the axis of perceived verticality. To ensure that

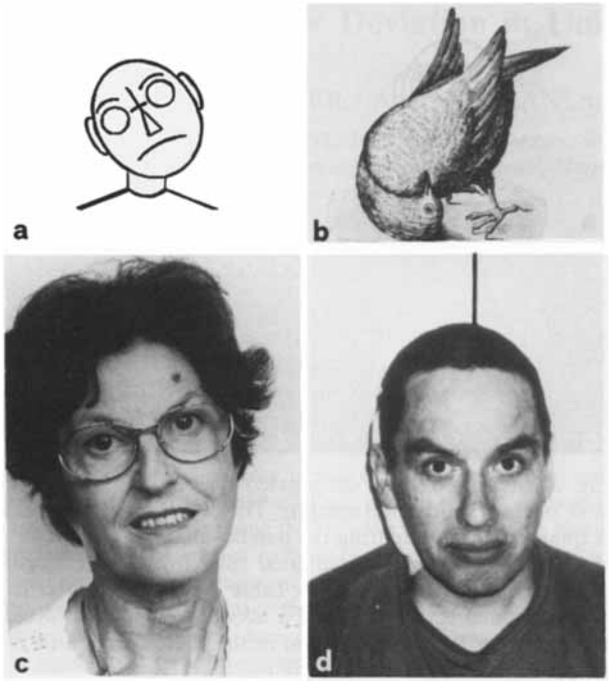

Fig. 4. Spectacular postural changes after peripheral unilateral destruction of the vestibular organ in a pigeon (*b*). Slight head tilt toward the operated side in two patients after right vestibular neurectomy (*c,d*).

the frame is horizontal, a spirit level was added to the transverse bar of the Maddox rod (16). The visual vertical is measured separately for each eye, on a graduated scale situated around the glasses (Fig. 2). Each test is performed four times, and the mean value is taken as the result.

Measurement of conjugated cyclotorsion of the eyes with ocular fundus photographs. The degree of cyclotorsion is quantified using photographs of the ocular fundus, taken with the head in an upright position. The papillo-macular axis tilt is measured, and is compared with the horizontal by means of a graduated ruler (Fig. 3).

Evaluation of head tilt. In patients following surgery, the head posture was tilted toward the operated ear, this was documented with portrait photographs in several patients (Fig. 4).

Additional investigations. All patients also underwent an extensive oto-neurological examination, including a clinical vestibular examination, a pure-tone audiogram, and an impedancemetry with measurements of stapedial reflex thresholds between 500 and 4,000 Hz and of reflex decay at 1,000 Hz. We recorded brainstem auditory potentials evoked by clicks (100 μs). Using computerized electro-nystagmography, we recorded spontaneous, positional and, optokinetic nystagmus, and measured saccades and smooth pursuit. We also performed bithermal caloric testing (using water at 44°C and 30°C, and iced water) and rotatory pendular testing, followed by recording of the fixation suppression of the vestibular nystagmus. A neurological check-up and neuro-radiological examinations (CT, MRI) were also performed.

In surgical patients, static visual vertical measurements were performed before oto-neurological surgery, a few days after surgery, and several weeks and months later. In non-surgical patients, measurements were performed a few days or weeks after the peripheral vestibular loss (13). In most patients, measurements were repeated several times at regular intervals during a one-year follow-up.

RESULTS

In surgical patients, the pre-operative otoneurological examination showed a symmetrical response to both caloric and pendular rotatory tests.

In the acute stage after the deficit, all surgical patients, as well as the patients suffering from an idiopathic peripheral vestibular deficit, showed the typical clinical picture of a unilateral peripheral vestibular loss, with vertigo, neuro-vegetative symptoms, spontaneous nystagmus toward the unaffected ear, and an absence of nystagmic response to caloric testing on the affected ear. Neurological and neuroradiological examinations did not indicated any additional central nervous system disease.

Surgical patients complained of vertical diplopia immediately after the ablative vestibular procedures. In the Hess-Weiss test performed within the first few post-operative days (Table I), a skew deviation, with hypotropia of the eye ipsilateral to the operated ear, was shown in cases 6 and 10. In cases 2, 3, and 4, vertical diplopia was clearly described by the patients, but measurements of SVV had to be interrupted because of vomiting. No hypotropia was found in 8 patients. SVV measurements were found tilted toward the operated side by 4° to 20° when evaluated using the vertical frame, and by 5° to 15° using the Maddox rod; in the monocular method, the tilt was greater in the eye ipsilateral to the operated ear. Ocular fundus photographs showed a conjugated cyclotorsion directed toward the operated ear by 4° to 18° in the right eye, and by 2° to 15° in the left eye.

During follow-up, vertical diplopia disappeared within one to two days, and skew deviation measured by the Hess-Weiss test also disappeared within 3 days post-operatively. In contrast, conjugated cyclotorsion and tilt of the static visual vertical persisted for weeks to months, with gradual improvement, as shown in Table I.

Table I. *Follow-up of ocular tilt reaction after surgical unilateral peripheral vestibular deafferentation*

Patient Nr.	Sex	Age (y)	Vestibular lesions	Side	Date	SVV Vertical frame	SVV Maddox rod RE/LE	Hess-Weiss	Ocular fund. RE/LE
1	M	30	Lab.	R	pr-op	0°	0°/0°		4°ex/3.5°ex
					3d po	+9.25°	+12°/+13°	NH	14°ex/6°in
					2w po	+8°	+8.5°/+8.5°		
					6w po	+3.25°	+6°/+6.5°		10°ex/3°in
					11mo	+1°	+3°/+4°		
2	M	64	Lab.	R	pr-op	+3°	+2.25°/+0.25°		
					1d po	+5.25°	+5.25°/+5.5°	VD	
					3d po	+6.75°	+5.75°/+5.25°	NH	
					2mo	+4°	+4.75°/+4.75°		
3	F	41	Lab.	R	pr-op	+1.25°	+2.75°/+0.25°		4.5°ex/3°ex
					1d po	+6.25°	+9°/+7.25°	VD	
					6w po	+3.5°	+5.75°/+5°	NH	12°ex/5.5°in
					3mo	+2.5°	+4.5°/+1.25°		15°ex/5°in
4	F	42	Lab.	L	1d po	−8.5°	−9.25°/−5.75°	VD	3°ex/10°ex
					3mo	−5°	−2.5°/−1.75°	NH	5°ex/10°ex
5	F	21	Lab.	R	pr-op	+0.75°	−1.75°/−0.25°		3°ex/7°ex
					2d po	+6.75°	+8.25°/+7.25°	NH	
					5w po	+6.25°	+4.5°/+5°		10°ex/3°ex
6	F	43	VN	L	pr-op	+1.25°	+3°/−2.5°		3°ex/5°ex
					2d po			LH	
					5d po	−5.75°	−7.5°/−12.15°	NH	
					8d po	−5°	−4.5°/−9.5		15°in/15°ex
7	F	45	VN	L	pr-op	−1°	0°/−3.75°		5°ex/7°ex
					2d po	−19.5°	−15°/−15°	NH	
					1w po	−8.5°	−8.75°/−8.75°		10°in/20°ex
					4mo	−0.75°	−5°/−4°		5°ex/15°ex
8	F	40	VN	L	pr-op	−0.75°	+1.75°/+0.25°		5°ex/2°ex
					4d po	−6.75°	−6.25°/−6.25°	NH	7°in/14°ex
					6mo	−4.5°	−5.5°/−5.25°		
9	M	45	VN	L	pr-op	+1.25°	+4.5°/+4.5°		7°ex/6°ex
					2d po	−1.5°	+2°/+0.25°	NH	
					6d po	−2°	+4°/+1°		3°ex/14°ex
10	M	50	VN	R	pr-op	+2°	+2.25°/+0.75°		9°ex/5°ex
					1d po	+4°	+6.25°/+2.25°	RH	15°ex/3°ex
					6w po	+1°	+3°/+0.25°	NH	
11	F	57	VN	R	pr-op	+3.75°	+4.25°/−3°		7°ex/7°ex
					4d po	+6.25°	+5°/+6°		
					2mo	+4.25°	+5.5°/+4°		
12	M	41	VN	R	pr-op	−0.75°	0°/−1°		7°ex/3°ex
					5d po	+6.75°	+5°/+5°	NH	17°ex/9°in
					2mo	+1.75°	+3°/+2°		
13	F	32	VN	L	pr-op	−0.25°	+4.5°/+1°		6°ex/10°ex
					8d po	−6°	−4.75°/−9.25°	NH	9°in/25°ex
					2mo	−3.5°	−3.5°/−6.5°		5°ex/15°ex

(y): years; SVV: static visual vertical measurement; Lab.: labyrinthectomy; VN: vestibular neurectomy; R.E/L.E..: right eye/left eye; +: tilt rightward; −: tilt leftward; VD: vertical diplopia; NH: no hypotropia; LH: left hypotropia; RH: right hypotropia; pr-op: pre-operative; po: post-operative: d: day; w: week; mo: month; ex: excyclotorsion; in: incyclotorsion

Among the patients with idiopathic unilateral vestibular neuritis and peripheral cochleo-vestibular loss, vertical diplopia was a prominent symptom in only two (cases 1 and 5, Table II). In contrast, among all patients, the Hess-Weiss test showed hypotropia in the eye ipsilateral to the affected ear in 5 patients, and in the eye contralateral to the affected in one patient (Table II). In the 4 tested patients (cases 3 to 6), the Maddox rod test showed a tilt of the SVV by 7° to 8°, while ocular fundus photographs showed

Table II. *Ocular tilt reaction in idiopathic unilateral peripheral vestibular lesions*

Patient Nr.	Sex	Age	Vestibular lesions	Side	Day ater onset	SVV Vertical frame	SVV Maddox rod RE/LE	SVV Hess-Weiss	Ocular fund. RE/LE
1	M	27	Neuritis	R	3			LH	
					14			NH	
2	M	52	Neuritis	R	14			RH	
					18			NH	
3	M	60	Neuritis	L	7		−7°/−5°	LH	0°/4°ex
					20		−7°/−5°	NH	
					40		−5°/−3°	NH	
4	M	65	Neuritis	R	14		+7°/+5°	RH	7°ex/2°in
					31		+3°/+5°	NH	
5	F	71	Neuritis	R	4		+8°/+8°	RH	15°ex/0°
					8		+6°/+6°	NH	
6	F	57	cochleo-vest. loss	R	2			RH	
					7			NH	
					30	+5°	+8°/+4°		13°ex/2°in
					60	+5°	+7°/+5°		
					360	+5°	+8°/+5°		13°ex/3°in

See Table I for abbreviations

4° to 15° of excyclotorsion in the eye ipsilateral to the affected ear.

In the non-surgical patients, vertical diplopia disappeared within a few days of the onset of peripheral vestibular loss. In contrast, the SVV monocular or binocular test showed a varying degree of tilt of the SVV, which persisted for many weeks to months. In one patient with sudden peripheral cochleo-vestibular loss (case 6, Table II), tilt of the SVV remained with no improvement for up to one year.

In the 32 normal subjects, static visual vertical measurements showed between +2° and −2° of tilt when evaluated with the vertical frame test, and between +4° and −4° of tilt with the modified Maddox rod test; positive and negative values indicate right and left deviation, respectively. Ocular fundus photographs performed in 24 of the normal subjects showed a slight (≤5°) excyclotorsion of both eyes in 19 subjects, and a combinaison of incyclotorsion (≤3°) of the right eye and excyclotorsion (≤5°) of the left eye in 5 subjects.

DISCUSSION

For more than a century, a spectacular eye-head posture reaction known as the ocular tilt reaction (OTR), including skew deviation and head tilt toward the lesion, has been described in animals in the acute stage after destruction of one vestibular organ (17, 18). In man, skew deviation is seen mainly in brainstem lesions (2–4). Since the development of oto-neurological surgery in recent years, this phenomenon has also been recognized after vestibular neurectomy and labyrinthectomy (5–8, 19, 20). In such cases, skew deviation has also been considered as part of the so-called ocular tilt reaction. This phenomenon is less marked in man than in animals (Fig. 4).

In our patients with unilateral peripheral vestibular deafferentation following ablative surgery, vertical diplopia due to skew deviation was reported immediately after surgery. The association of this vertical form of strabismus with the other signs of OTR, including deviation in the visual vertical, head tilt, and conjugated cyclotorsion directed toward the operated side corroborated our previous observations (8, 9) as well as those of other authors (10). Post-operatively, the various signs of OTR disappeared or decreased progressively in a typical chronological order. Vertical diplopia, as a clinical symptom, was present only in the first few days after surgery, and sometimes only for a few hours. Skew deviation disappears within a few days. In contrast, static visual tilt and conjugated cyclotorsion persisted for weeks or months. The Maddox rod test and ocular fundus photographs showed greater tilt and cyclotorsion in the eye ipsilateral to the peripheral vestibular lesion. A comparision of values using the vertical frame test (binocular method) and the Maddox rod test (monocular method) showed that results from the former method tended to return more quickly to normal, suggesting that binocular input might allow a better compensation. However, other parts of the central nervous system could also play a role in subjective vertical perception. Indeed, in one of our surgical patients (case 9) we found no tilt of SVV, even though conjugated cyclotorsion was clearly deviated toward the operated ear. In the post-operative SVV measurement, this patient, who worked as carpenter, first indicated that his SVV tilted by 8° to the side of the operated ear, but, during the same evaluation procedure, was able progressively to reajust the deviated pointer to the vertical, with a final error of only 2°. This reajustment of the SVV may have been made possible by his cognitive knowledge of the vertical and his perception of angles, as used daily in his professional activity.

In patients with vestibular neuritis, the symptom of vertical diplopia and the measured skew deviation, at the beginning of the clinical picture, initially suggested a central nervous system lesion. However, the absence of associated neurological symptoms, and the fact that an extensive neurological investigation was negative, favored the hypothesis of the peripheral vestibular disorder. In the literature, various visual phenomena have been noted to occur in acute idiopathic unilateral peripheral vestibular deficit. These include an impression of rotation, oscillopsia due to spontaneous nystagmus and difficulty in focusing (21). It is possible that difficulty in focusing was related to an unrecognized ocular tilt phenomenon. Indeed, in our patients we found a significant tilt of the SVV toward the side of the affected ear in more than 40% of patients with a peripheral vestibular or cochleo-vestibular loss (unpublished data). However, because of the delay between the acute phenomenon and the otoneurological investigation, skew deviation was rarely observed.

After surgical deafferentation, the eye-head postural changes and the deviation of vertical perception were observed extending over weeks to months. In non-surgical patients, the phenomenon was also transient except in one patient (case 6), in whom cyclotorsion and subjective visual tilt perception persisted, and were almost unchanged one year after the onset of the acute cochleo-vestibular loss. This distinctive evolution suggests that central mechanisms of compensation in oculo-vestibular pathways may differ according to the extension or origin of the peripheral vestibular lesion in the vestibular organ and/or nerve. The mechanisms of OTR following peripheral unilateral vestibular lesions are presumably related to a lesion of the otolith organs and/or to changes in the afferent graviceptive pathways (6,10). OTR may often remain unrecognized in man, and can be masked by spontaneous nystagmus and marked neuro-vegetative symptoms.

Our observations indicate that vertical diplopia due to skew deviation, as part of OTR, may occur in patients with sudden peripheral vestibular lesions of surgical or non-surgical origin.

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