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Evaluation of Tullio Phenomenon by Computerized Dynamic Posturography

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INTRODUCTION

The phenomenon of sound-induced vestibular symptoms and signs has been known for a long time. It was labeled 60 years ago in light of Pietro Tullio's experiments [1–3]: after creating minuscule fenestrations in the horizontal semicircular canal he elicited pendular head movements and nystagmus in pigeons by leading the sound of a flute to their ear. Observing the motion of dye particles in the labyrinthine fluids, Tullio also observed that currents are created in the columbine semicircular canals in response to sound stimuli, with perfect correspondence between the frequencies of the applied sound and the oscillations of these internal currents. He showed that sound waves spread from perilymph to endolymph and stimulate the hair cells of the cristae ampullares.

Historically, the Tullio phenomenon (TP) was thought to be present only when a fistula existed in the bony labyrinth. This led later to the pathognomonic association between TP and congenital syphilis wherein a labyrinthine fistula explained the occurrence of sound-induced vestibular malfunction. As more cases of vestibular symptoms induced by intense sound were reported in noisy environments such as industry and aerospace, several researchers engaged in the study of the underlying mechanism [4–9] as well as the nystagmic eye movements produced in response to intense sound [4,10–12]. Later studies focused on the functional assessment of the acoustically stimulated vestibular system in terms of performance on force platform [10, 13–15]. Thus, during the last two decades, a correlation was found between TP and positive results on electronystagmography (ENG) and posturography in a variety of pathologic conditions of the middle and inner ear, such as chronic otitis media [11,13], congenital and noised-induced sensorineural hearing loss (NIHL) and hearing loss due to other causes [11,14,16], direct vestibular trauma [14,16], Menière's disease [10,13,16–18], and operated otosclerosis [11].

The presence of TP also was noted in normal individuals—guinea pigs, monkeys, and humans [19,20], and later even found as an ubiquitous finding in normal subjects [21]. It was argued, however, that the contrasting results in the latter study were due to methodological errors and are inconclusive [13]. Yet it does not imply that the sono-vestibular reaction does not occur in normal subjects [14], perhaps as a function of stimulus intensity and duration [22] i.e., above 120 dB SPL in the mid-frequency range [23].

Scanning the literature for the different stimuli used in different studies (Figure 1), one can notice that the sound pressure levels used to elicit a Tullio response in healthy subjects were higher than those used in subjects with pathologies of the ear.

The vestibular function tests with acoustic stimulation represent a different and more sensitive evaluation technique than the classical fistula sign performed with pressure levels many orders of magnitude greater [24].

The Tullio phenomenon has been studied only by nystagmographic methods and by static posturography, as mentioned earlier. We could not find any study of vestibular responsiveness to acoustic stimulation evaluated by computerized dynamic posturography (CDP) in the scrutinized literature. Static posturography falls short of revealing pure vestibular malfunction manifested clinically as standing or walking unsteadiness. Since in the study of the TP one is concerned exclusively with the vestibular contribution to the function, or dysfunction of equilibrium, the development of the CDP was opportunely to help us to pioneer the appropriate technique for investigating the interaction between balance and loud sound within the inner ear.

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MATERIALS AND METHODS

Computerized Dynamic Posturography

The occurrence of changes in stability when exposed to sound was carried out by applying an acoustic stimulus to the ears of a subject while striving to maintain balance on the platform of the computerized dynamic posturograph. In dynamic platform posturography computerized force vector analysis was added for the purpose of improvement to the older static platform posturography. The latter, also called stabilometry, constitutes a quantified Romberg test, evaluated with respect to changes in center-of-sway amplitude, distance, or velocity. The more recent dynamic posturography estimates body sway angles from vertical projections of the center of force. Nashner has carried this evaluation a step further: the Equitest® system, known generically as computerized dynamic posturography (CDP), attempts to ferret out the effects of various sensory inputs to the brain and to relate them to overall on-feet balance and stability [27–29]. It also uses the Romberg test with a fixed platform, but includes test conditions in which the platform and visual environment are moved to reduce the subject's ability to use visual and proprioceptive information for balance.

CDP constitutes a comprehensive sensorial function test of the equilibrium by virtue of its inherent feature of isolating the vestibular, visual, and somatosensory contribution to overall balance, unlike older posturography techniques that served as mere systemic balance tests.

By isolating the vestibular input, CDP constitutes a true vestibular function test, similar to ENG, with the specification that ENG is not designed for direct assessment of a patient's equilibrium, but simply monitors eye movements caused by the vestibulo-ocular reflex and only indirectly reflects vestibular impairment if it exists.

CDP employs a computer-controlled, menu-driven moveable platform called a dual force plate and a moveable background that surrounds the patient almost completely and fills the patient's visual field. The background contains colored patterns simulating an abstract

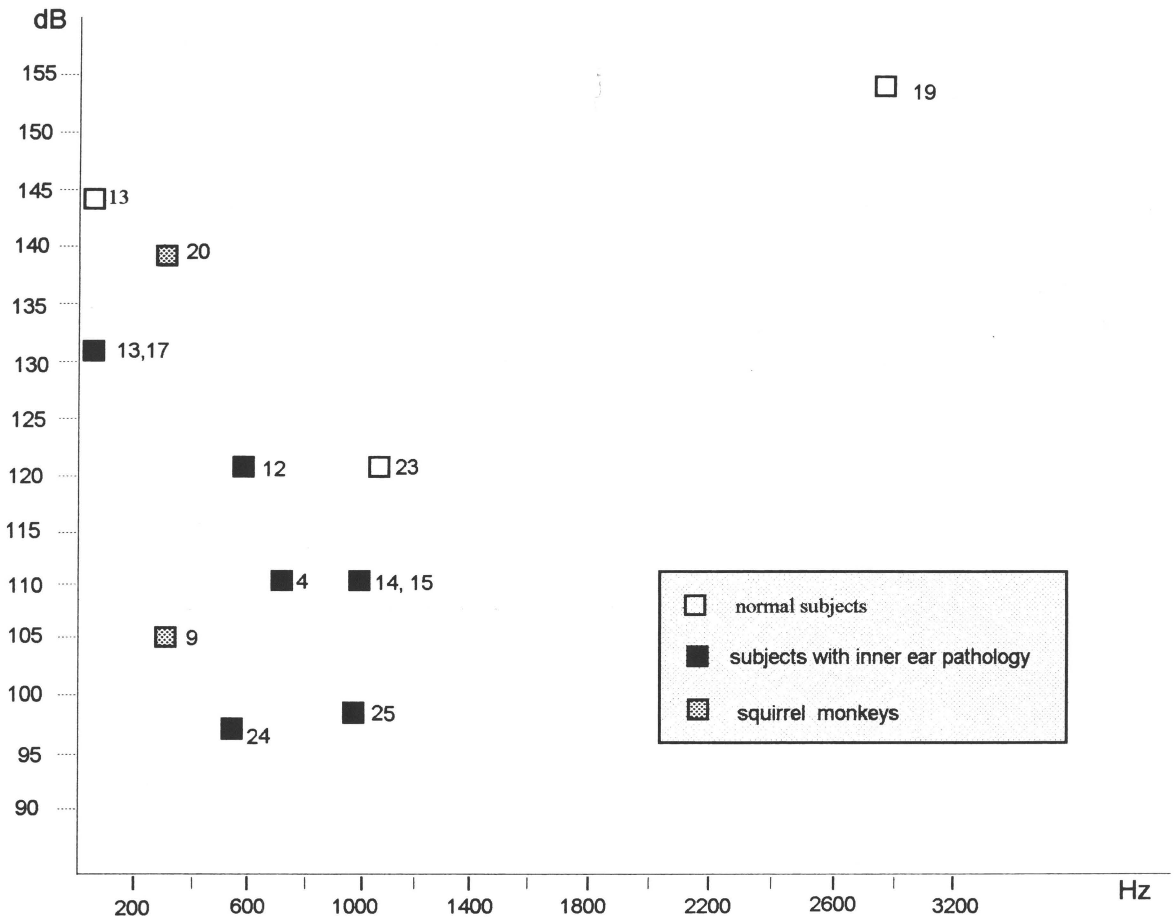

Figure 1. Most effective acoustic stimuli for evoking a Tullio response, as described in the literature. Numbers refer to the bibliographic source.

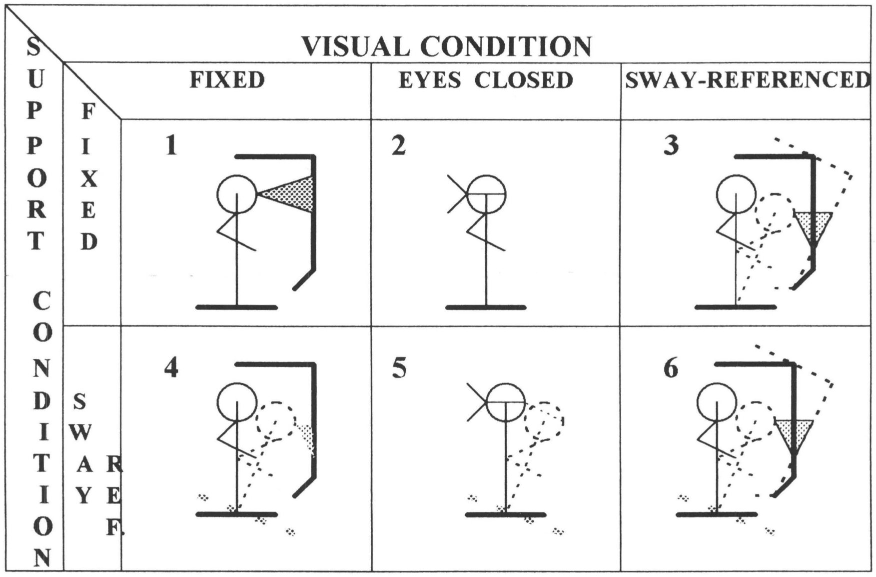

Figure 2. The six different sensory organization conditions of CDP.

landscape and provides the surface for visual orientation and fixation. Both the platform and background are "sway-referenced," that is, move along with the anteroposterior sway of the patient. The testing protocol requires that the patient stand on the platform with feet comfortably straddled shoulder-width apart. The body sway is monitored by pressure-sensitive gauges located in each quadrant of the platform. As the patient sways around her or his center of balance, the platform and the background track the patient's motion, providing an objective measure of the actual AP sway.

The **sensory organization test (SOT)** is the portion of the CDP that is most useful in the assessment of patients with suspected vestibular disorders. During testing, the computer monitors the forces generated by the patient on the platform, including vertical forces and horizontal plane (shear) forces.

The SOT uses six separate tests (conditions), lasting 20 seconds each (Figure 2).

- **SOT 1** is a quantified version of the Romberg test. The subject stands with eyes open while the visual surround and the ground are stable.
- **SOT 2** is equal to SOT 1, except that the eyes are closed.
- In **SOT 3**, performed with eyes open, the visual surround moves in response to the body sway (i.e., it is sway-referenced, thus providing distorted visual conditions).
- In **SOT 4,** the platform is sway-referenced (for somatosensory distortion) and the surround stable.
- In **SOT 5**, with eyes closed, the platform is sway-referenced.
- In **SOT 6** both the platform and the surround are sway-referenced.

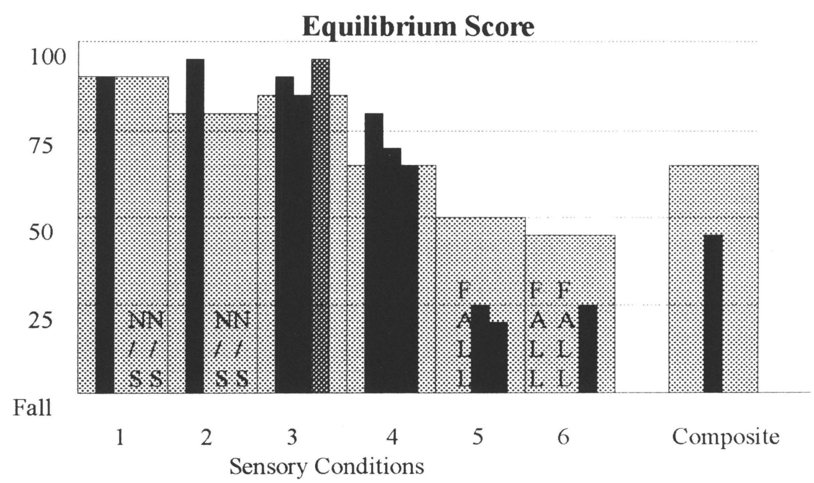

Figure 3. Example of the individual and composite scores obtained from an abnormal SOT in a patient with vestibular dysfunction. The abnormal scores are indicated by the bars falling within the shaded area.

From each test an ***equilibrium score*** is computed (Figure 3). It is based on the assumption that a normal individual can exhibit anterior to posterior sway over a total range of 12.5 degrees (6.25° anterior, 6.25° posterior) without losing balance. The equilibrium score is calculated by comparing the angular difference between the patient's calculated maximum anterior to posterior displacements of the center of gravity to this theoretical maximum displacement. The result is expressed as a percentage between 0 and 100, with 0 indicating sway exceeding the limits of stability and 100 indicating perfect stability. The composite equilibrium score is calculated by averaging the scores for each condition.

When the composite score falls within the abnormal range, the second interpretation test is to identify the sensory dysfunction that contributed to the overall sensory organization abnormality. This process is called sensory analysis (Figure 4). This is accomplished by computing sensory ratios between the average equilibrium scores on specific pairs of sensory test conditions (Table 1).

Age-matched limits of normality are shown as shaded areas on the test result printout and the abnormal scores are indicated by bars falling within these areas (see Figures 3, 4).

Patients

Three groups of subjects were included in the study (Table 2).

- **(A)** A study group of patients suffering from NIHL, and who complained of dizziness when exposed to sound i.e., sono-vestibular symptoms (Tullio phenomenon).
- **(B)** A second group comprising patients with NIHL without a history of Tullio phenomenon.

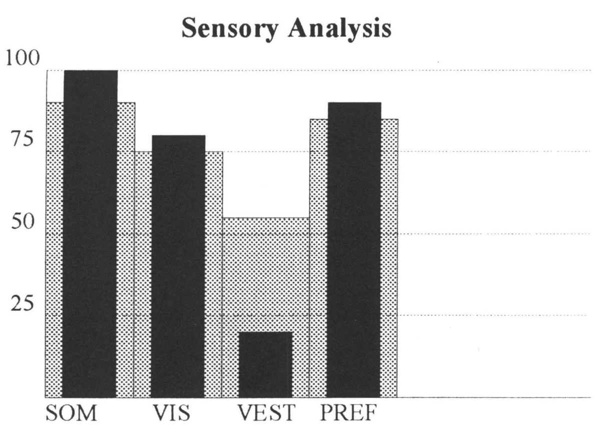

Figure 4. Example of the sensory analysis based on the scores obtained from the same patient (fig. 3).

Table 1. Sensory Analysis: Calculation and Significance of the Sensory Ratios

Somatosensory	Cond. 2/Cond. 1	The ability to use input from the *somatosensory system* to maintain balance
Visual	Cond. 4/Cond. 1	The ability to use input from the *visual system* to maintain balance
Vestibular	Cond. 5/Cond. 1	The ability to use input from the *vestibular system* to maintain balance
Preference	Cond. 3 + 6/ Cond. 2 + 5	Degree to which patient relies on visual information to maintain balance, even when that information is incorrect

The NIHL in these first two groups was established and documented by history, physical examination, and audiometry. All patients with abnormal visual, audiovestibular, and neurologic function, except NIHL (group B) and NIHL accompanied by Tullio symptoms (group A), were excluded from the study.

- **(C)** A control group of subjects with normal hearing, matched with the study groups in all other aspects.

Informed consent was obtained from the subjects, whereas the experimental protocol was approved by the local Ethics (Helsinki) Committee and the Israeli Ministry of Health.

The differences in ages of the participants in the three study groups were not significant when compared statistically.

Testing protocol: CDP with acoustic stimulation

The SOT was performed three times 15 minutes apart: 1) in the regular mode (quiet stance), 2) quiet stance in

Table 2. Study Subjects in Computerized Dynamic Posturography with Acoustic Stimulation (Preliminary Stage)

Study Group	No. of Subjects	Age (yrs) Average	SD
A: NIHL + sonovestibular symptoms (Tullio)	8 (M:F = 7:1)	48.375	14.19
		$p > 0.57$ (N.S.)	
B: NIHL	12 (M:F = 10:2)	51.75	10.86
		$p > 0.46$ (N.S.)	
C: Normal hearing (controls)	15 (M:F = 12:3)	48.26	13.37

SD = standard deviation; NIHL = noise-induced sensorineural hearing loss; M:F = male-female ratio; N.S. = not significant.

SOTs 1, 2, 3, and 4, followed by acoustic stimulation in SOT 5 and SOT 6 (i.e., in the "vestibular conditions"). 3) quiet stance again.

A pure-tone sound of 110 dB at 1000 Hz generated by a portable audiometer was delivered in continuous mode. From the wide range of acoustic stimuli used to provoke a sonovestibular response, we favored one of the lowest intensities that efficiently, yet still safely, elicits a Tullio response. This sound pressure level is well within safety limits, in accordance with the Illinois Occupational Diseases Act [30] and the Occupational Safety and Health Administration Regulation [31]. Bearing in mind that acoustic stimuli in unusual environmental conditions always affect both ears, we preferred to employ binaural stimulation in an attempt to prove the hypothesized correlations in accordance with the hazards of quotidian reality.

RESULTS

The results are summarized in Figures 5 and 6.

First, the composite score obtained in the three study groups was analyzed. Since the composite score is an average of all sensory conditions, it reflects the overall on-feet stability of the subject. Not unexpectedly, a remarkable drop in the composite score of the patients in group A was witnessed ($p < .002$, by t-test: two-sample, assuming equal variances). This instability continued even after the stimulus was delivered during the second test, in the third trial 20 minutes later. However, this decrease was not significant. The interesting finding is that these patients showed a worse balance than groups B and C from the start on the first test, before any stimulus was applied ($p < .01$ and $p < .006$, respectively, by t-test: two-sample, assuming unequal variances). The normal subjects (group C) behaved almost similarly to those with NIHL without Tullio symptoms (group B), i.e., they showed a slight (not significant) improvement during the sequence of the three consecutive sensory organization trials with the second carried out under acoustic stimulation.

Since in the study of the Tullio phenomenon we were interested in both the overall equilibrium, as well as the vestibular system in particular, the vestibular ratio given by the CDP software was investigated next. In group A, the vestibular component of the equilibrium demonstrates again a decrease upon stimulation with sound (see Figure 6). This decrease contrasted once more with the results in the other two groups, namely B

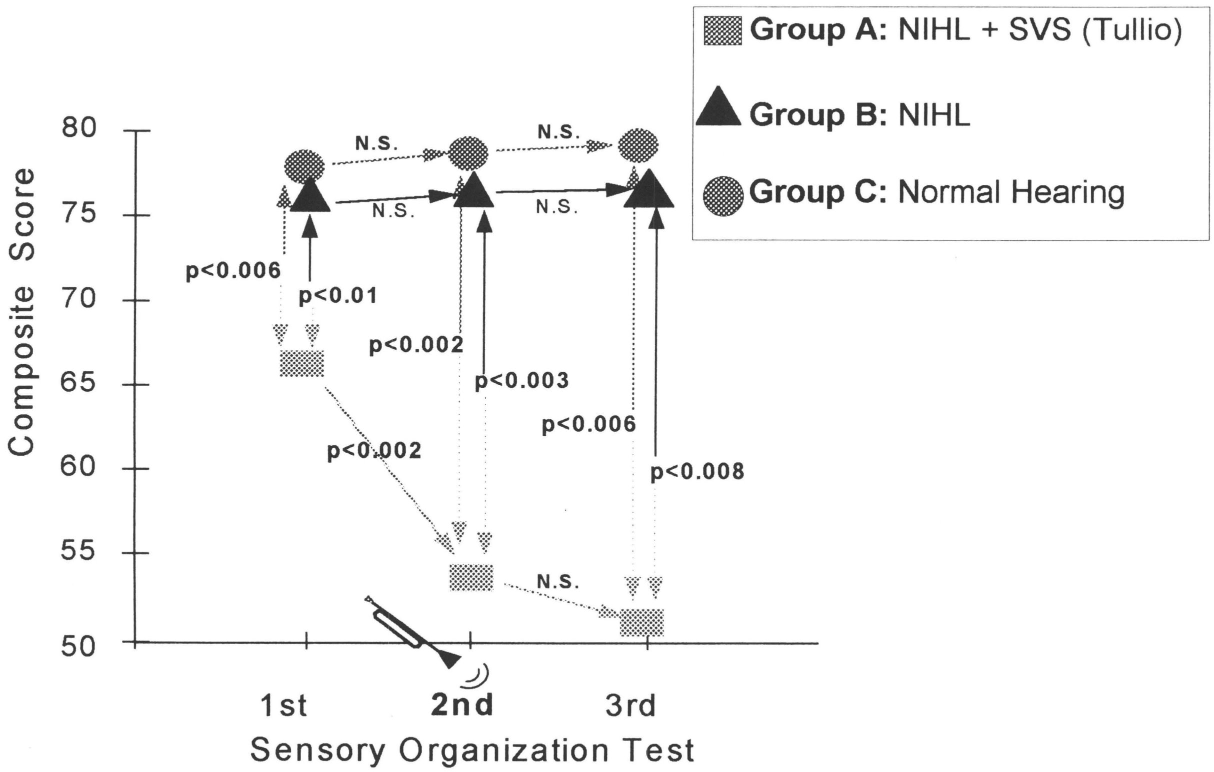

Figure 5. Composite score (overall equilibrium) in three consecutive sensory organization tests, with the second carried out partly under acoustic stimulation.

and C: they marked an increase in both the composite score and the vestibular ratio upon repeated tests. Nevertheless, this increase was not significant.

DISCUSSION

The current study focused on the postural stability of normal subjects and patients with NIHL, as well as on patients with NIHL accompanied by a history of sonovestibular symptoms. The postural responses were monitored on the Equitest (i.e., the CDP), in three consecutive SOTs, with the second performed partially under binaural delivery of sound (110 dB at 1,000 Hz).

When comparing our results to those found in previous works in which static posturography methods were used, we cannot but pay heed to the controversy surrounding the subject in the literature.

Hadj-Djilani [14,15] studied the performance on force plate under sound-stimulation with the exact same sound stimulus we delivered in our work (1000 Hz,110 dB SPL). This was applied to patients with Menière's disease, chronic otitis media with vertigo, and uni- and bilateral vestibular lesions. The normal subjects in this study showed unresponsiveness or a destabilizing response; a stabilizing response was encountered in evolutive ear diseases like chronic labyrinthitis, otosclerosis, Menière's disease, familial and autoimmune hearing loss.

The results obtained by Ishizaki, Pyykko & Aalto [13,17] were not simply different, but they showed a surprisingly sharp contrast with the findings. In this work they studied Tullio phenomenon using low-frequency sound (25–63 Hz, 130 dB SPL) and observing the change in stability on a static posturograph: not only did the intense sound have no deleterious effect on the balance of normal subjects, but they improved their postural stability during sound stimulation, probably through the alerting response. On the other hand, patients with Menière's disease, peripheral vestibular dysfunction, and chronic otitis media with vertigo showed increased body sway, causing instability.

Our normal subjects, as well as the patients with NIHL without a medical history of Tullio symptoms, showed no significant change in postural stability, and in this our results resemble those obtained by Ishizaki et al.

The positive medical history of sonovestibular symptoms was confirmed objectively by CDP with

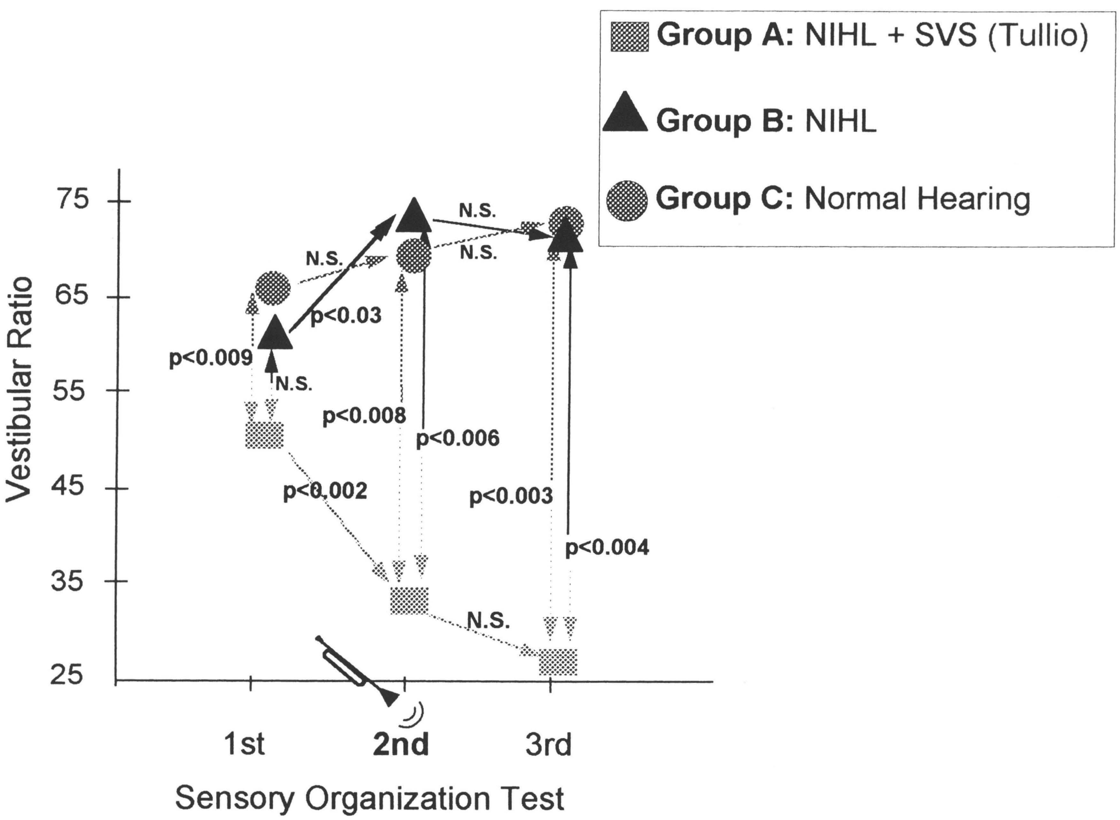

Figure 6. Vestibular ratios obtained in CDP in three consecutive sensory organization tests, the second under stimulation with sound. NIHL = noise-induced sensorineural hearing loss; SVS = sonovestibular symptoms.

sound stimulation with a high statistical significance. This establishes the described method as a sensitive testing technique for validating the existence of Tullio phenomenon in patients who exhibit noise-induced hearing loss NIHL, even when employing stimuli of harmless intensities (110 dB) and short durations ($6 \times$ 20 sec). This stimulus is well within the noise exposure standards and therefore safe and a temporary threshold shift is the only unwanted effect that it produces. Therefore, it can be routinely used for detecting this disturbing and potentially dangerous phenomenon in patients with NIHL who complain of sudden dizziness, or give protean descriptions of feeling bad when they happen to be exposed to intense noise in a concert hall, discotheque, airfield, or at work.

Patients with NIHL accompanied by sono-vestibular symptoms showed a more deficient equilibrium and profound vestibular impairment, even in the absence of noise, despite having been matched to the other two study groups in all conceivable aspects (age, gender, audiovestibular history, and audiological findings). The significance of this finding has yet to be interpreted.

The diagnosis of vestibular malfunction related to sound exposure may be of crucial clinical significance. In NIHL, we should keep in mind the possibility of a currently asymptomatic vestibulopathy that is progressing to a disturbing vertigo, independently of the cochlear pathology. Three sensory systems are involved in the maintenance of balance: vestibular, visual, and somatosensory. When vestibular damage occurred and remains undiagnosed, the overall function of balance relies only on the other two systems left. When, in addition, other sudden sensory deprivation occurs, or if new relationships develop between the sensory systems involved in balance, spatial disorientation may transpire under these circumstances. Exposure to intense noise in unusual environmental settings during flying under changing gravity, diving, physical activity in darkness or night driving, might produce conditions that in certain circumstances could even endanger life. Virtually all patients who suffered phonal trauma or chronic exposure to noise (e.g., aviation employees, industry and army personnel) and complain of sono-vestibular symptoms should be tested for the presence of Tullio phenomenon by means of computerized dynamic posturography with acoustical stimulation for an objective corroboration of their complaint before continuing activity in a noisy environment, thus preventing dangerous loss of balance when exposed to noise.

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