Unusual Tullio phenomena

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THE Tullio phenomenon consists of the production of vestibular symptoms and/or signs by an acoustic stimulus (Tullio, 1926; Huizinga, 1934, 1935). The effect depends upon the functional integrity of the vestibular labyrinth only, and is reproduced in animals when the cochlea has been ablated. Bleeker and de Vries (1949) have shown that a microphonic (analogous to the cochlear microphonic) can be recorded from the crista of the fenestrated semicircular canal of the pigeon and the frequency response curve of this cristal microphonic parallels the frequency response curve for the Tullio phenomenon. This vestibular microphonic can be recorded from any of the vestibular end-organs (Huizinga *et al.*, 1951) and it has been observed in fishes (Pumphrey, 1939), reptiles and amphibians (Adrian *et al.*, 1938).

It is generally considered that the Tullio phenomenon occurs in man only when more than one mobile window opens into the internal ear on the vestibular side of the vestibular membrane (Cawthorne, 1956). An apparent exception to this rule was reported by Land in 1957. Although Land's 60-year-old male complained of 'giddiness' and a pulsating tinnitus when he lay on his left side and an acoustic stimulus could produce pallor, sweating and a falling reaction, Mitchell (1957) suggested that a psychological factor might be involved in that case. No otoscopic or audiometric abnormality was detected in that case, and an acoustic stimulus was not observed to produce nystagmus. Thus we considered that three other exceptions to the rule, but with objective findings, were worthy of reporting.

Case Reports

Case 1.

A 28-year-old male window dresser (G.S.) was referred to the Neuro-otological Clinic of the Royal National Throat, Nose and Ear Hospital on 10 April 1968 complaining of 'buzzing' noises in the right ear together with recurrent episodes of subjective, swaying vertigo over the previous two years. The vertiginous episodes were of a few seconds' duration and were sometimes, but not always, precipitated by the loud, rumbling noises of trucks. The episodes were not associated with nausea, vomiting, or changes in hearing. The patient considered that his hearing was normal, although on direct questioning at a later stage of the examination he said that there was some difficulty on the right side.

The patient was married with no children. He had two sibs (both females)

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of whom one was older and the other younger than he was. There was no previous history of otological disorders in the patient or in his immediate family, nor any history of allergic or psychosomatic disorders in the patient, but his mother suffered from asthma and one of his sibs suffers from hay fever. This patient smoked 20 cigarettes daily and drank two pints of beer each week.

Apart from some hyperaemia of the larynx there was no visible abnormality in the nose or throat. Both tympanic membranes were observed to be imperforate and mobile. The Rinne test was positive on either side and a Weber test was not lateralized. Sweep frequency Békésy audiometry showed a normal threshold in the left ear but a 50 db. hearing level[1] for low tones in the right ear (Fig. 1). This low frequency hearing loss was subsequently confirmed by evoked response audiometry. The comfortable loudness level for a 1,000 Hz tone was recorded in each ear by Békésy audiometry and the measured levels on each side were seen to be within 10 db. of one another (Fig. 1). Fixed frequency

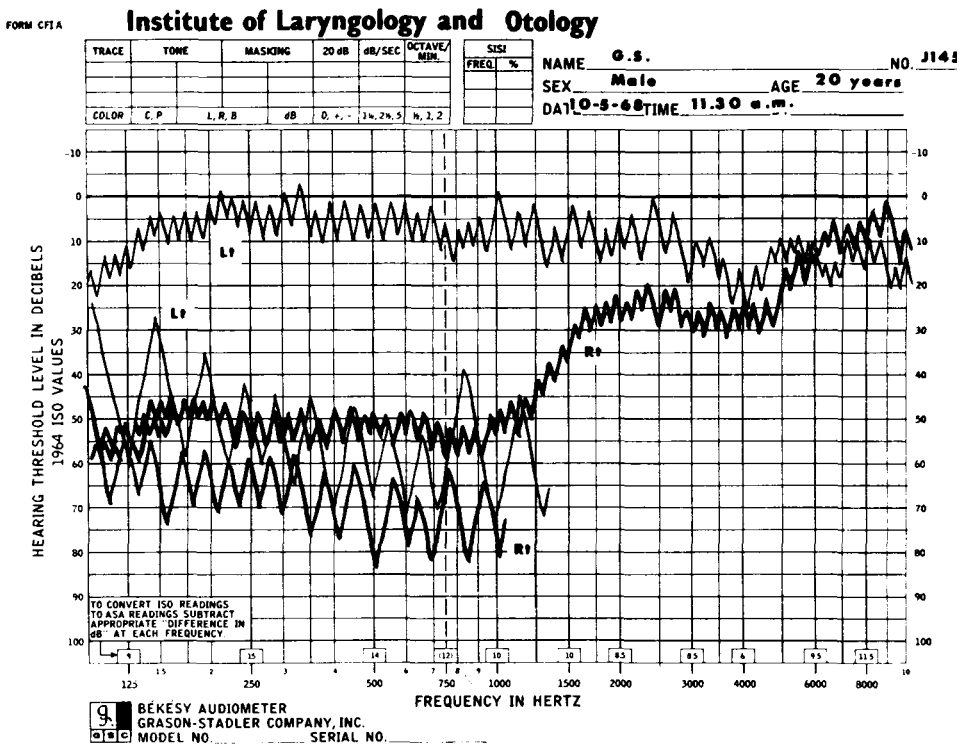


FIG. 1.

Sweep frequency Békésy audiogram of G.S. at time when he was experiencing Tullio phenomenon.

[1] All hearing measurements in this study were made with audiometers calibrated to ISO recommendations.

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Békésy audiometry showed no threshold drift. Alternative binaural loudness matching at 1,000 Hz showed equal loudness in the two ears for a tone of 100 db. HL. At 1,000 Hz, the SISI score was 100 per cent in the right ear. Speech audiometry showed no discrimination loss. Using the Békésy audiometer, the tinnitus in the right ear was matched to a 750 Hz 15 db. SL tone in the left ear. Klockhoff and Lindblom's (1966) glycerol test produced a 10 db. shift in the auditory threshold at 250 Hz in the right ear.

With the eyes closed and the feet together, the patient could stand with no undue sway for not less than 30 seconds, and the heel-to-toe test was performed normally. There was no clinical spontaneous or positional nystagmus and the optomotor nystagmus showed no directional preponderance. Examination with electro-oculography confirmed the absence of spontaneous nystagmus and also failed to elicit nystagmus by means of the fistula test. Thermal vestibulometry, using ampullopetal stimuli, showed a normal response (Fig. 2). The sono-ocular test (presentation of acoustic stimuli with an audiometer and electro-oculographic recording of eye responses with the eyes closed) was positive. Transient nystagmus was produced with tones of 500 Hz and 1,000 Hz at 110 db. HL and with audiometer masking noise at 90 db. presented to the right ear (Fig. 3). The patient identified the sensation produced by these sounds as identical with that occurring during the vertiginous episodes. This acoustically-induced nystagmus was habituatable.

Examination of the other cranial nerves, including the corneal reflexes, showed no abnormality. Personality tests showed a psychosomatic-V profile (Fig. 4) for the MMPI (Minnesota Multiphasic Personality Inventory), and the MPI (Maudsley Personality Inventory) gave a neuroticism score of 36 and an extraversion score of 4, whilst the Alpert-Haber test gave a score of 7, i.e. no significant test anxiety.

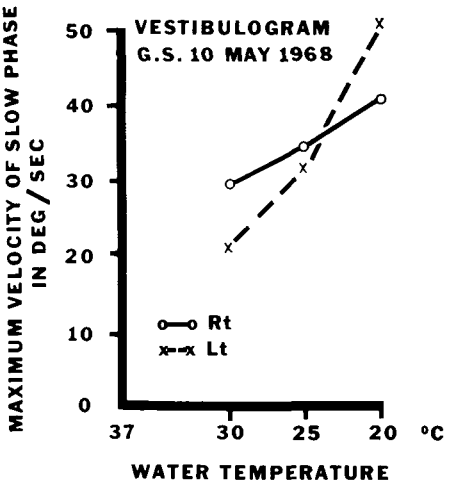


FIG. 2.

Thermal vestibulogram of G.S. with strength of nystagmus as a function of the temperature of water used for aural irrigation. Patient in prone position.

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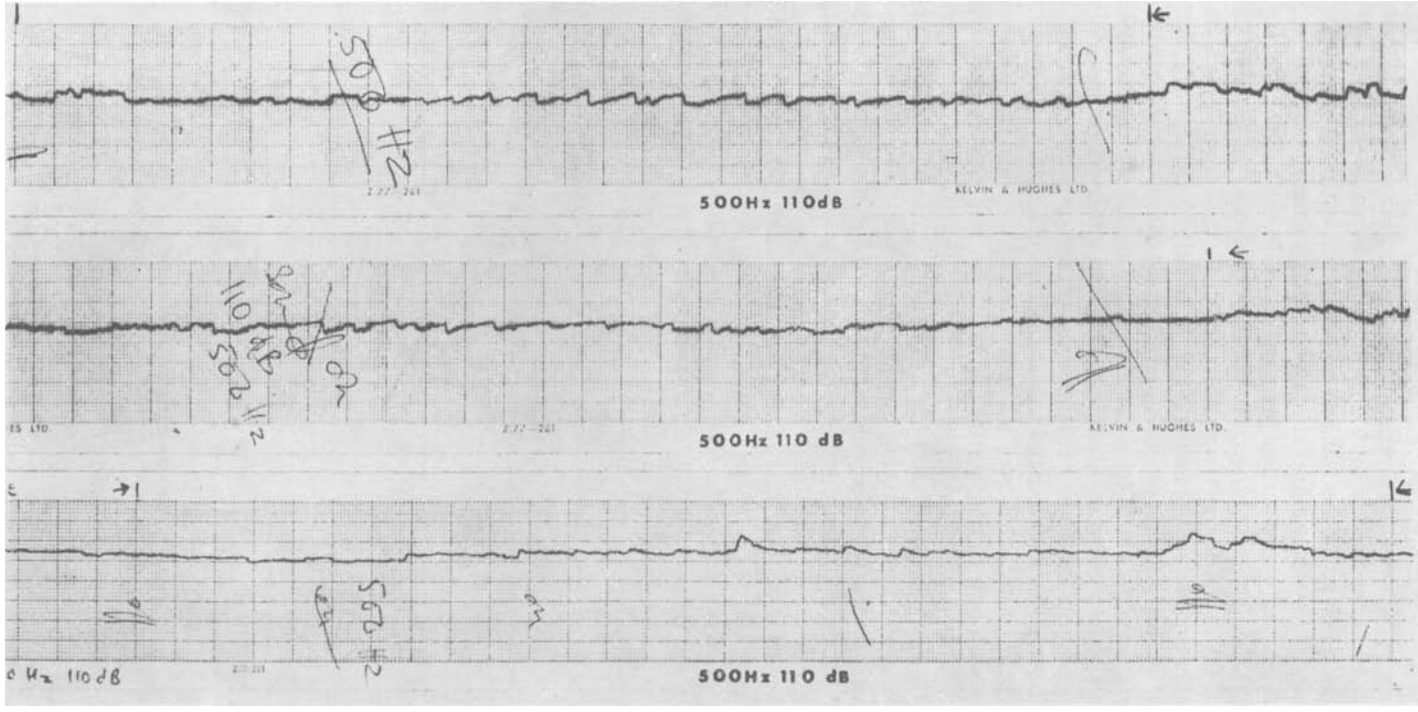


FIG. 3.

Sono-EOG of the two cases of endolymphatic hydrops. The top two tracings show vestibular nystagmus to the left induced in G.S. by a tone of 500 Hz at 110 db. HL presented to the right ear. The bottom tracing shows vestibular nystagmus to the right induced in A.L. by a 500 Hz 110 db. HL tone presented to the right ear.

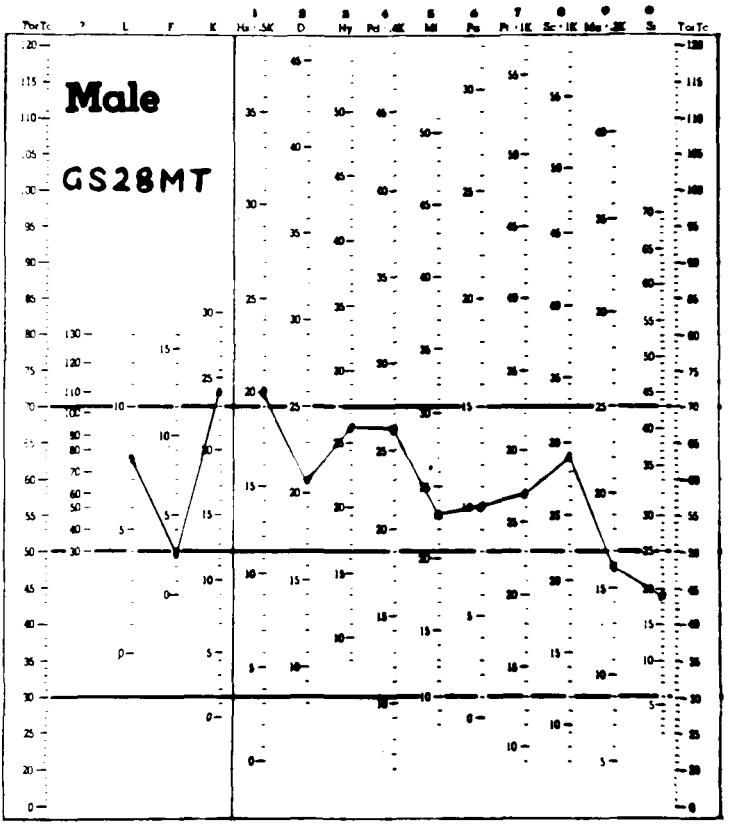


FIG. 4.
MMPI (Minnesota Multiphasic Personality Inventory) of G.S. showing typical psychosomatic-V pattern (on scales Hs, D and Hy).

The blood pressure was 124/80, examination for treponemal antibodies was negative and X-rays of the internal acoustic meatuses showed no abnormality.

The patient was reassured and the condition explained to him. No medication was prescribed nor any operation advised.

When the patient was seen again on 21 November 1968, we learned that he had had no further episodes of vertigo nor of the Tullio phenomenon since we had first seen him and his hearing loss had improved by some 20 db. (Fig. 5).

Case 2.

A 55-year-old Prison Officer (A.L.) was referred to the Neuro-otological Clinic of the Royal National Throat, Nose and Ear Hospital on 29 October 1968 with the complaint of recurrent episodes of objective, rotary vertigo over the previous seven years. The episodes of vertigo lasted anything from about a second to half an hour and were sometimes associated with nausea and vomiting but never with loss of consciousness or changes in his auditory symptoms.

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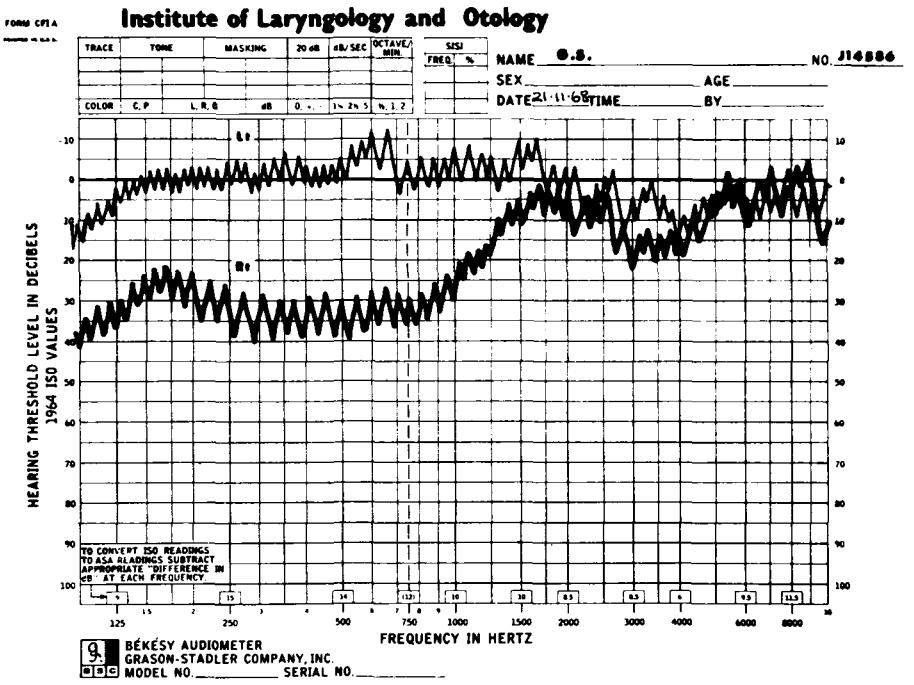


FIG. 5.

Follow-up Békésy audiogram of G.S. at a time when he was no longer experiencing the Tullio phenomenon.

He also said: 'when a heavy lorry goes by, especially when I am walking under a bridge, it causes dizziness and makes me shake my head.' For the past seven years, he had also had some difficulty in hearing, especially under noisy conditions, and a continuous hissing sound in the right ear. For the past five years there had also been an intermittent sensation of fullness in the right ear. The vertigo was ameliorated by prochlorperazine.

From 1930–1937, this man had served in the Regular Army, then in a Colonial Police Force and, from 1939–1945, in the Tank Corps. During his service with the latter organization, he was, on one occasion, thrown against a tank by bomb blast and was rendered unconscious for two minutes. He had been a rifle instructor and had fired 'thousands' of rounds of 0·303 ammunition, sustaining temporary tinnitus on occasions but remembering no hearing loss. He had never worn ear protectors. He had received quinine in 1932 for the treatment of malaria but did not recollect any tinnitus or impairment of hearing. The patient smoked 28 cigarettes a day and drank 10 pints of beer and half a bottle of whisky each week.

The patient, a single man, was from a family of seven children of whom the eldest sister (now dead) had suffered from asthma and the youngest sister from severe headaches and she had had a 'nervous breakdown'.

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Examination showed no visible abnormality in the ears, nose or throat. A forced whisper was heard at a distance of 50 cm. on the left side, but not at all on the right side. The conversational voice was heard at a distance of 5 cm. on the right side. The Rinne test was positive on either side and the Weber test was not lateralized. Sweep-frequency Békésy audiometry showed a hearing loss for frequencies in excess of 3,000 Hz in the left ear, with a maximum loss at about 4,500 Hz. There was a hearing loss for all frequencies in the right ear, the level being of the order of 50 db. for the low frequencies (see Fig. 6). Fixed frequency Békésy audiometry showed no threshold drift and comfortable loudness levels recorded for a pulsed test tone at 1,000 Hz showed complete loudness recruitment at 70 db. HL (Fig. 7). Loudness recruitment was also indicated by the Fowler test and, when the test was being performed, the patient spontaneously reported diplacusis binauralis dysharmonica. Speech audiometry showed that the discrimination loss in the right ear was not in excess of 16 per cent. Using the Békésy audiometer, the tinnitus in the right ear was matched to a 4,000 Hz 22 db. SL tone in the left ear.

With the eyes closed and the feet together, he could stand with no undue

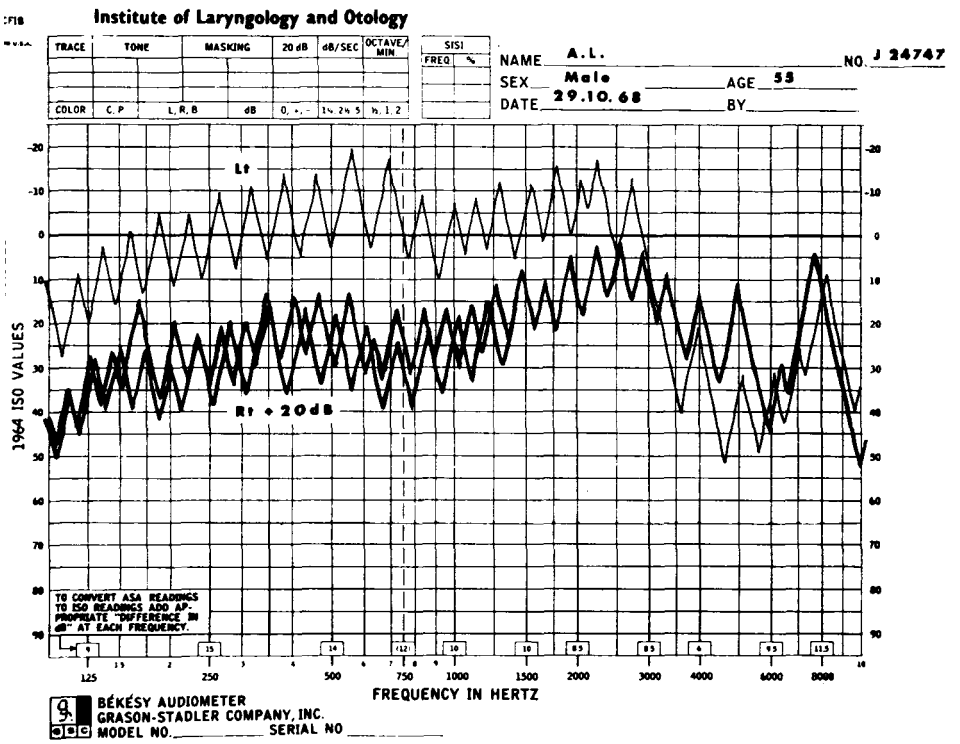


FIG. 6.

Sweep-frequency Békésy audiogram for A.L. when he was first seen and was found to have a Tullio phenomenon. Note that the hearing levels for the threshold in the right ear are '+ 20 db.'.

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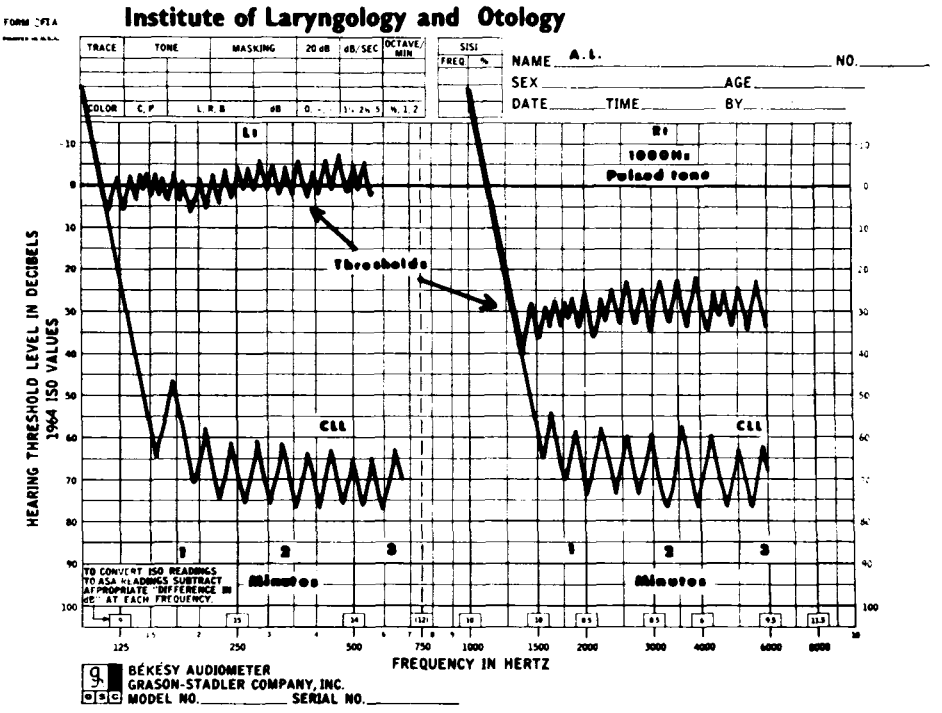


FIG. 7.

Comfortable loudness levels for A.L. recorded with a Békésy audiometer and using a pulsed test tone. Note that both CLLs are about 70 db. HL in each ear, indicating the occurrence of complete loudness recruitment.

sway for not less than 30 seconds, but with the heel-to-toe test he took eight steps before losing his balance. He was slightly unsteady with chair circling, but equilibrium defence was good and there was no clinical spontaneous or positional nystagmus. The optomotor nystagmus showed no directional preponderance. Examination with electro-oculography confirmed the absence of spontaneous nystagmus and also failed to elicit nystagmus by means of the fistula test. The caloric test showed results within normal limits. The sono-ocular test was positive. Transient nystagmus was produced with a tone of 500 Hz at 110 db. HL presented to the right ear (Fig. 3). The nystagmus was directed towards the right but on the first occasion only that the sound stimulus was applied.

Examination of the other cranial nerves, including the corneal reflexes showed no abnormality. The MMPI (Fig. 8) showed a profile that Sullivan and Welsh (1952) have said is indicative of a psychosomatic personality. The score on the Alpert-Haber test was 11.

The blood pressure was 146/90, examination for treponemal antibodies was negative and X-rays of the internal acoustic meatuses showed no abnormality.

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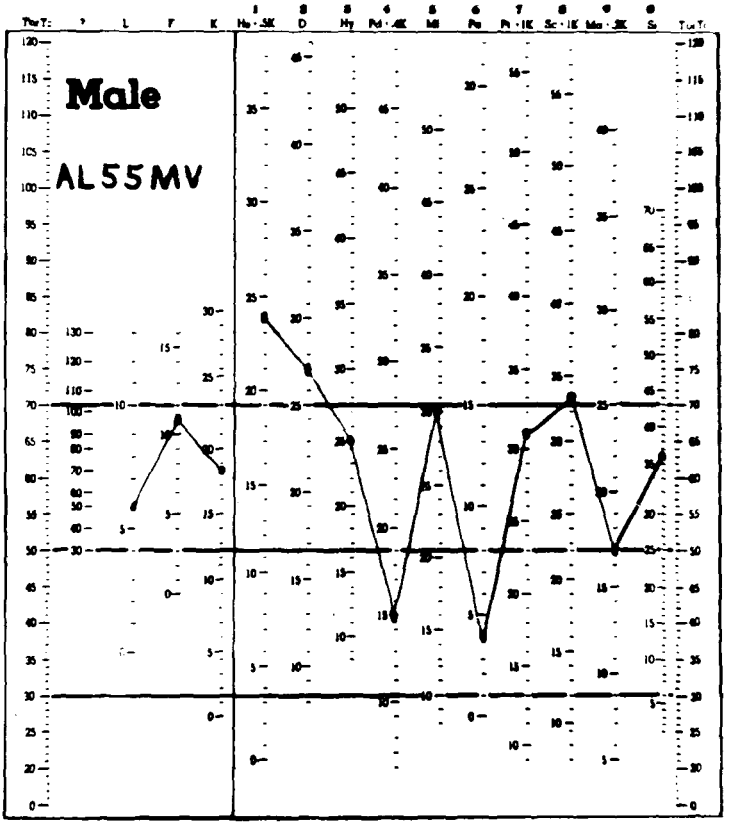


FIG. 8.

MMPI for A.L. Note elevation of profile on neurotic triad (Hs, D and Hy) with Hs> D> Hy.

When the patient was seen again on 17 February 1969 he was no longer experiencing the Tullio phenomenon and the sono-ocular test was negative. A Békésy audiogram made on this occasion is shown in Fig. 9.

Case 3.

A 27-year-old male civil engineer (R.K.G.) presented at the Vertigo Clinic of the All-India Institute of Medical Sciences (New Delhi) on 14 January 1969. He complained of objective, swaying vertigo, associated with nausea and vomiting, following injury by a pencil to the left ear four months previously. Difficulty in hearing and a continuous low pitched tinnitus in the left ear had also followed the injury. Moreover, the patient complained that loud sounds produced a feeling of imbalance associated with 'shock' in the left ear. This particular symptom he had noticed in the crowded bazaar.

There was nothing relevant in the patient's previous history or in that of his family.

Apart from a deviated nasal septum to the left, there was no visible abnormality in the nose or throat. The right tympanic membrane appeared normal

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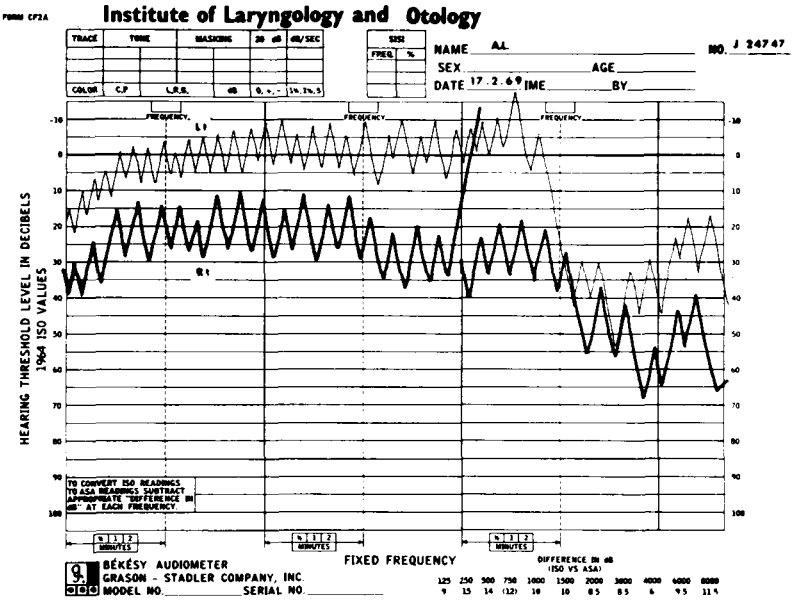


FIG. 9.

Follow-up sweep frequency Békésy audiogram on A.L. at a time when he was no longer experiencing the Tullio phenomenon.

but there was a scar in the posterior half of the left tympanic membrane. The Rinne test was positive on the right but not on the left. Manual audiometry showed there to be a 70 db. hearing level on the left side, with normal thresholds on the right. At 1,000 Hz in the left ear, there was an air-bone gap of 20 db. and the SISI score was 100 per cent.

With the eyes closed and the feet together, he could stand with no undue sway for not less than 30 seconds and the heel-to-toe test showed no abnormality. There was no clinical spontaneous or positional nystagmus but examination with electro-oculography with the eyes closed showed a vestibular nystagmus to the right. The caloric test was within the limits of normality. A Tullio phenomenon was produced by clinical testing. When the patient's eyes were open, noise from a Bárány noise box applied to the left ear produced a vestibular nystagmus to the right. The induced nystagmus was associated with a feeling of transient unsteadiness. Both the induced nystagmus and the vertigo were habituatable.

Exploration of the left middle ear by one of us (S.K.K.) on 17 January 1969 showed no evidence for a fistula. There was, however, a subluxated, hypermobile stapes the base of which had been pushed into the vestibule.

Remarks

In view of the history of recurrent episodes of vertigo (some of which were not precipitated by loud noises), unilateral tinnitus and an ipsilateral

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low frequency recruiting type hearing loss in the absence of otoscopic or relevant radiological or serological abnormalities, a diagnosis of Ménière's disorder was made in the first two cases. In each of these cases, this diagnosis was supported by the psychosomatic-type personality profiles and, in the first of these cases, by the 10 db. shift in auditory threshold for frequencies around 250 Hz with Klockhoff's glycerol test. In the third case, the operative findings showed that the condition was due to direct trauma dislocating the stapedo-vestibular joint, associated with rupture of the annular ligament and subluxation of the stapes base into the vestibule.

The ability to record nystagmus in the first two cases, to observe it in the third case and to reproduce these results with acoustic stimuli applied to the involved ear shows that these cases are undoubted examples of the Tullio phenomenon. The most notable feature, however, is that there is no evidence for the presence of a second mobile window or of a fistula (other than via the fenestra vestibuli in Case 3) opening into the vestibular labyrinth in any of the three cases.

It is tempting to try to arrive at some unifying concept which might provide a basis for a hypothesis to explain the Tullio phenomenon in these three cases. First, it is likely that the existence of an ipsilateral endolymphatic hydrops in the first two cases is more than coincidental. Secondly, the operative findings in the third case showed that the stapes base was almost certainly in contact with the saccule. Now it is known that, in endolymphatic hydrops, the distended saccule may be in intimate contact with the stapes base (Cody *et al.*, 1967). Thus the factor common to these three cases may be the contiguity of the tympano-ossicular chain with the membranous labyrinth, with no intervention by perilymph as in the normal state. The precise vestibular end-organ that is being stimulated in these cases is a matter of conjecture. Although vestibular nystagmus is commonly associated with stimulation of the cristae, there is evidence that stimulation of the otolith mechanisms may modify an existing nystagmus (Owada and Shiizu, 1960; Owada *et al.*, 1960).

The presence of normal caloric responses in the two endolymphatic hydrops cases would support the hypothesis that, whilst one requires a sufficient degree of hydrops to bring the saccule into contact with the stapes base, the condition must not be sufficiently advanced to have resulted in a non-functioning vestibular mechanism. An adequately sensitive vestibular apparatus is essential for the occurrence of the phenomenon.

The spontaneous disappearance of the Tullio phenomenon in the first two cases may be held to be due to a diminution of the hydrops and consequent loss of contact between the membranous labyrinth and the stapes. This hypothesis is supported by an associated improvement of the low frequency threshold by the order of 20 db. in both of these cases. Fluctuations of the auditory threshold in endolymphatic hydrops are attributable to variations in endolymphatic pressure (Tonndorf, 1957).

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It is now over a century since Ménière described the disorder that now bears his name, so it is surprising that the occurrence of the Tullio phenomenon has not been previously observed. It is possible that this is because the symptom is not sought and, when it is reported, it is confused with phonophobia. Moreover, it is possible that the phenomenon never occurs to a sufficient degree to be observed without recourse to electro-oculography, a technique which is still so little used in examination of patients with Ménière's disorder.

Summary

The Tullio phenomenon is reported in three cases where there was no evidence for the existence of more than one mobile window opening into the internal ear on the vestibular side of the vestibular membrane. One of these cases was due to traumatic dislocation of the stapedo-vestibular joint with subluxation of the stapes base into the vestibule. The other two cases were diagnosed as endolymphatic hydrops. The hypothesis was entertained that, in all three cases, the phenomenon was due to direct contact of the stapes base with the membranous labyrinth.

Spontaneous remissions occurred in the two cases of endolymphatic hydrops.

Acknowledgements

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