QUANTITATIVE MEASUREMENTS OF EYE MOVEMENTS IN A PATIENT WITH TULLIO PHENOMENON

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Abstract — The Tullio phenomenon consists of vestibular symptoms on exposure to high-intensity acoustic stimuli, reflecting pathological stimulation of semicircular canals or otoliths. We report a patient with posttraumatic Tullio phenomenon to illustrate how precise measurement of eye movements during auditory stimulation, using the magnetic search coil technique, may characterize movements that are not clinically apparent or easily measured by other means. Such measurements in patients with surgically verified lesions may further elucidate the mechanisms responsible for this phenomenon.

Keywords — Tullio phenomenon; nystagmus; perilymph fistula; magnetic search coil.

Introduction

The Tullio phenomenon refers to sound-induced vestibular symptoms (1). Although prior papers have reported clinical, audiological, and posturographical findings, documentation of eye movements has been hindered by the limitations of electro-oculography (2), because the movements are often small and may be predominantly torsional (that is, rotations around the line of sight). The magnetic search coil technique allows precise measurement of eye and head rotations in all three planes, and is now quite widely available (3). We used this technique to study a patient with posttraumatic Tullio phenomenon and present data to illustrate how it might help to determine whether the disturbance is more typical of dysfunction of semicircular canals or otoliths. Such information might ultimately help in planning treatment.

Patient and Methods

Case report

A 36-year-old woman developed Tullio phenomenon following head trauma. One year prior to our evaluation, she had been hit by an automatic door and fell, apparently without losing consciousness; she sustained bilateral jaw dislocation. She had not experienced dizziness prior to the head injury, but afterwards reported that “my eyes move” whenever her phone rang. This feeling would start immediately when the phone started to ring and stopped as the ringing stopped. When this happened, she sometimes felt a little unsteady, but did not experience rotatory vertigo or oscillopsia. She also reported sometimes feeling unsteady when she made fast head movements. Her prior medical history was remarkable for a congenital, familial hearing deficit that had not been specifically diagnosed. Since childhood she had worn hearing aids in both ears that provided satisfactory improvement of her hearing. She had not noticed any additional hearing loss since the accident. By alternately removing each of her hearing aids she was able to identify her right ear as respon-
possible for the problem: the symptoms only occurred when the right hearing aid was in place. She was unable to provoke the phenomenon by the Valsalva maneuver or by applying manual pressure to her external auditory canal. Clinical examination showed a nystagmus with horizontal and torsional components that was present exclusively during the time that her telephone was ringing. It was low-amplitude, variable, and sometimes could only be detected with an ophthalmoscope. There was no noticeable head or body tilt during acoustic stimulation. To date, our patient has not undergone surgical exploration of her middle ear, but intends to consider this if her symptoms persist.

Eye Movement Measurements. The patient gave informed consent according to the Helsinki Declaration of 1975. Horizontal, vertical, and torsional rotations of both eyes and of the head were recorded using the magnetic search coil technique (3,4). Search coils were calibrated prior to the experimental session using a protractor device. Data were filtered (bandwidth 0 to 100 Hz) prior to digitization at 300 Hz (for routine ocular motor testing) or 1000 Hz (for recordings with acoustic stimulus). For determinations of the latency to onset of nystagmus, the sound of the phone was recorded through a microphone and digitized without filtering at a sampling rate of 1000 Hz. In order to determine the frequency components, recordings of the sound were performed at sampling rates of 3000 and 5000 Hz, and the main frequencies were evaluated using a Fast Fourier transform. The visual stimulus consisted of a red laser spot that was rear-projected, under the control of mirror galvanometers (General Scanning CX 660), onto a semitranslucent tangent screen at a viewing distance of 1.2 m.

Experimental Stimuli and Data Analysis

1. Routine ocular motor testing. We tested re-fixation saccades as the target made ± 10° horizontal and vertical jumps. Smooth pursuit was tested as the target (laser spot) moved sinusoidally through ± 20° at 0.3 Hz in either the horizontal or vertical direction. The visually enhanced vestibulo-ocular reflex (VVOR) and convergence were also tested. Data analysis was performed using interactive programs written in the ASYST language. Gain (ratio of eye movement and target movement) of saccadic amplitude, smooth pursuit, and VVOR velocity were calculated (4,5). Peak velocities of saccades, convergence angle, and slow-phase velocities of nystagmus were measured.

2. Recordings with acoustic stimulation. To standardize the sound stimulus, we first used sound frequencies of 125, 250, 500, 750, 1000, 1500, 2000, 3000, 4000, 6000, and 8000 Hz through a hearing testing device (Maico portable audiometer). During this testing with headphones, the patient did not wear her hearing aids. Sound pressures up to 105 dB HL were applied and, probably due to the severity of hearing loss, none elicited the Tullio phenomenon. Equipment was not available for accurate sound field testing. Therefore, we tested the patient while she wore her hearing aids and used her telephone, which reliably induced the Tullio phenomenon. The telephone was placed 0.4 m from the patient, and it was determined that it had no effect on the search-coil signal. Each phone ring lasted about 2 s. We measured the effect of the telephone ring on the stability of gaze during fixation of the stationary laser dot and in complete darkness as the patient attempted to look at the remembered target location. We also applied acoustic stimulation during smooth pursuit and VVOR testing.

Results

1. Routine Ocular Motor Testing

Saccadic gain was 0.90 horizontally and 0.98 vertically. Saccadic amplitude/peak-velocity relationships were normal. Smooth pursuit tracking gain was 0.97 horizontally and 0.96 vertically. The gain of the VVOR was 1.04 horizontally and 1.07 vertically. The patient was able to converge 21° in response to a near stimulus. These results are all within the normal range (4–6). However, during all conditions except fixation of a stationary target, there was a low-ampli-
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tude, torsional nystagmus, with quick phases beating clockwise with respect to the patient. This nystagmus was variable, being maximal during smooth pursuit when slow phase velocity was 1.6°/s and beating frequency was 3 Hz. Attempted fixation in complete darkness revealed, in addition to the torsional nystagmus, variable horizontal left-beating and vertical down-beating components. The horizontal and vertical slow phase velocity reached a maximum of 1.2°/s, and the frequency sometimes reached 2 Hz. She was unaware of this spontaneous nystagmus.

2. Recordings with acoustic stimulation

The sound stimulation reliably provoked a horizontal right-beating and a torsional clockwise beating nystagmus; an example is shown in Figure 1. We applied a total of 22 sound stimuli during fixation paradigms (9 with a target and 13 in complete darkness), and nystagmus was triggered every time; the mean latency to onset was 16 ms. This response was the same during fixation of a stationary target and during attempted fixation in darkness. There was no effect on the vertical component, that is, during fixation there was no vertical eye movement, and in darkness the spontaneous downbeat nystagmus (if present) continued. The average slow phase velocity was 1.1°/s (max. 1.5°/s) horizontally and 0.8°/s (max. 1.3°/s) torsionally; the average frequency of quick phases was 0.6 Hz.

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**Figure 1.** Tullio phenomenon during fixation of a stationary target. As soon as the acoustic stimulation starts, conjugate horizontal right-beating and torsional clockwise-beating nystagmus commenced. Note the absence of any spontaneous nystagmus prior to, and of vertical nystagmus during, this sound stimulation. The single position traces are offset for convenience of display; upward deflections indicate rightward (horizontal), upward (vertical), or clockwise (torsional) eye rotations, with respect to the patient. The sound signal is only displayed for timing information. Its record is distorted by the aliasing effects of a sampling rate at 1000 Hz, well below the required Nyquist frequency for this signal.

**Figure 2.** Tullio phenomenon during attempted fixation in complete darkness. Horizontal position signal of the right eye is shown. Conventions are the same as those in Figure 1. (A) Horizontal left-beating spontaneous nystagmus takes place in darkness without acoustic stimulation. At the beginning of this recording, sound stimulation had just ended and, at the end, a new stimulation had begun. Note that direction of the slow phase reverses each time. (B) Two seconds of sound stimulation in darkness. As soon as the sound starts, the slow phase movement reverses direction from rightwards to leftwards, and the nystagmus is now beating to the right. As the sound stops the direction of the movement again immediately reverses.
Note that the horizontal left-beating spontaneous nystagmus (present only in darkness) changed its direction to right-beating during acoustic stimulation (Figure 2).

Sound stimulation had no influence on the performance of smooth pursuit (gain horizontal 0.98, vertical 0.97) or VVOR (gain horizontal 1.06, vertical 1.07). No head movements were induced by the acoustic stimulation.

Frequency analysis of the telephone ring revealed a frequency spectrum from 750 to 1200 Hz with 2 peaks, at 905 and at 1140 Hz.

Discussion

Tullio mainly studied the phenomenon that carries his name in pigeons (1); his experiments have been repeated and extended by others (7,8). If a small opening is made into the vestibule of the bony labyrinth, a stereotyped response to acoustic stimuli occurs, consisting of a contralateral head tilt; this is interpreted as being due to utricular stimulation. An opening made into one of the bony canals leads to head nystagmus in the plane of the affected canal; for the lateral canal, quick phases of head nystagmus are directed towards the side of the acoustic stimulation. This type of head nystagmus is the way that a pigeon responds to stimulation of the crista. Huizinga (7) suggested that the artificial opening in the bony labyrinth has the same effect with regard to sound waves as the round window has to the cochlea: it allows the sound waves, which otherwise would have been extinguished through interference, to escape from the labyrinthine fluid. In this case, fluid movement stimulates not only the cochlea, but also the crista of the affected canal.

Prior clinical studies also suggest otolithic and semicircular-canal variants of Tullio phenomenon, although the mechanism is often in doubt. The otolithic type has been well characterized in a patient studied by Dieterich and colleagues (9), who showed a tonic, torsional, and vertical (skew) eye deviation, head tilt, postural imbalance, and a subjective tilt of the visual environment (the ocular tilt reaction). An initial phasic vertical and torsional deviation of the eyes occurred with a latency of about 20 ms, and was followed by a smaller tonic effect that lasted as long as the sound stimulation. Surgical exploration in this patient showed a subluxated stapes footplate with a hypertrophic stapedius muscle, causing pathologically large movements of the stapes footplate during the sound-induced stapedius reflex. Thus, the saccule or utricle may have been directly stimulated by the stapes footplate. Other patients who have been thought to have an otolithic type of Tullio phenomenon have shown either tonic torsional eye movements with head tilt (10), conjugate vertical eye movements (11), or horizontal nystagmus (12). Patients with Tullio phenomenon attributed to pathological stimulation of a semicircular canal have been less clearly defined (9,13,14).

In our patient, sound stimuli induced a horizontal right-beating and torsional clockwise-beating nystagmus (the horizontal component was more pronounced) with a latency similar to that of the vestibulo-ocular reflex (16 m) (15). When she was not maintaining steady fixation—either during other eye movements or when in darkness—there was a low-amplitude nystagmus beating leftward, clockwise, and downward; this suggested a small vestibular imbalance. During sound stimulation, the horizontal nystagmus reversed its direction, but the torsional (and vertical) nystagmus did not. There was no vertical (skew) eye movement or head tilt that might suggest otolithic stimulation. Thus, we postulate that the reversal of horizontal nystagmus was due to sound stimulation of the crista of the right horizontal semicircular canal, which overcame a component of an underlying vestibular imbalance. Our patient had congenital hearing loss—which is associated with Tullio phenomenon (16)—but only developed symptoms after head trauma, suggesting a fistula as the cause, although this was not surgically confirmed.

In many reported patients with Tullio phenomenon, it is difficult to be sure of the underlying mechanism; this problem often arises because the eye movements have not been well characterized. We have shown that it is possible to make precise measurements of 3-dimensional eye and head movements using the search coil technique, which is now available at most teaching centers. This procedure is safe, well
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tolerated, and is the standard by which other methods are judged (17). In patients such as ours, it allows reliable characterization of eye movements that are small and may remain undetected by clinical examination. Better characterization of the eye movements in Tullio phenomenon in patients in whom the underlying disorder has been surgically confirmed may elucidate the mechanisms by which the symptoms and signs arise.

Addendum

We were able to obtain an audiogram subsequent to measurement of eye movements. This confirmed a bilateral sensorineural hearing loss, moderate-to-severe (loss ranging 50 dbHL at 200 Hz to 85 dbHL at 8000 Hz) in the left ear and severe-to-profound (loss ranging 60 dbHL at 250 Hz to 100 dbHL at 8000 Hz) in the right ear. Impedance audiometry demonstrated normal tympanic membrane mobility but absent stapedial reflexes, bilaterally.

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