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Case Report

Vestibular Atelectasis: Decoding Pressure and Sound-Induced Nystagmus With Bilateral Vestibulopathy

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We present the case of a 27-year-old male who presented with vertigo when pressing the entrance of his right auditory meatus and exposing his right ear to loud noise. A diagnostic procedure revealed bilateral labyrinth weakness, which was confirmed by caloric and rotational testing. The ocular vestibular evoked myogenic potentials investigation demonstrated a significant weakness of the right utriculus, whereas the cervical vestibular evoked myogenic potentials were normal, indicating preservation of the saccular response. Radiologic studies did not show evidence of labyrinthine dehiscence. We suspect the newly described association of this clinical syndrome with the previously described histopathology of vestibular atelectasis accounts for these findings.

Key Words: Vestibular atelectasis, bilateral vestibulopathy, sound- and pressure-induced nystagmus, vestibular diagnostic.

INTRODUCTION

The presence of bilateral vestibulopathy with Tullio’s phenomenon or a positive fistula test with normal radiological imaging may be explained by the collapse of the endolymphatic space in the labyrinth, known as vestibular atelectasis.

CASE REPORT

A 27-year-old male patient was admitted to our outpatient clinic. He complained of vertigo when driving a car and changing altitude pressure when flying and diving, and also by pressing the entrance to his right auditory meatus or exposing his right ear to loud sound. Recently, he experienced a severe attack of vertigo after a rapid head movement, as if someone pushed him to the right side. He noticed that by pressing on the lower part of his auditory meatus with his index finger, horizontal ocular movements to the right were produced, whereas the pressure on the central or upper part of auditory meatus caused a move to the opposite (left) side. He noted oscillations of his visual field when walking, consistent with oscillopsia. As a child, he experienced head trauma and concussion. His hobby is diving, and he practices it regularly. His hearing was good, with occasional tinnitus in his right ear. He did not have any significant balance disorders before.

The patient was adequately informed about the methods and objectives of this study. He has voluntarily accepted to participate in the study. Written informed consent was obtained. Otoscopy was normal. The patient was swaying more than normal to the left and right after 10 seconds during the Romberg test on foam rubber with his eyes closed. The fistula test was positive in the right ear. When applying pressure with his index finger to the lower part of the auditory meatus, right directional horizontal nystagmus appeared, and pressing on the central or upper part resulted in left directional horizontal nystagmus (see Supporting Information, Video 1, in the online version of this article). A head impulse test was positive bilaterally, but much more to the right, which suggested a bilateral lesion of the vestibulo-ocular reflex (VOR), much more pronounced on the right. The Fitzgerald-Hallpike caloric test within videonystagmography confirmed bilateral, but equal, vestibular weakness with vestibular sensitivity below 5°/sec (Fig. 1). The video head impulse test demonstrated reduced VOR gain of

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0.16 on the right side, whereas on the left side it was 0.54, with overt and covert corrective saccades bilaterally, confirming a significant bilateral vestibular lesion (Fig. 2). Whereas the cervical vestibular evoked myogenic potentials finding was normal, the ocular vestibular evoked myogenic potentials findings showed a significantly higher peak-to-peak amplitude to the left, with the amplitude ratio up to 47% (Fig. 3). In the rotational test, it was not possible to calculate the result, as both labyrinths were practically nonexcitable. The patient’s average air-conduction pure-tone hearing threshold was 8 dB, with a word recognition score of 90%. Impedance audiometry
showed a bilaterally normal type A record, with the stapedius reflex present both ipsilaterally and contralaterally. High-resolution computed tomography imaging of the temporal bones was normal (Fig. 4), whereas magnetic resonance imaging showed physiological appearance of the structures of the inner ear, internal acoustic meatus, and cranial nerves in the posterior cranial fossa, as well as standard signal intensities in the mastoid region, middle ear, perilabyrinthine, and labyrinth area (Fig. 5).

**DISCUSSION**

The Italian physiologist Pietro Tullio described sound-induced vertigo in 1929.2 In his honor, the phenomenon of sound acting on the vestibular sense, which is one of the most prominent symptoms of superior canal dehiscence syndrome, is called the Tullio phenomenon. Bilateral vestibular hypofunction (BVH) is relatively common in the adult German population (3.6%–5.2%).3–5 Clinical presentation includes balance disorders observed as instability and insecurity while walking, especially in a dark environment and on uneven surfaces, because the deficit of vestibular sense is compensated by signals from the visual and somatosensory senses. The dominant symptoms are ocular, such as oscillopsia and visual blurring (25%–50% of those affected), which appear by rapid head movements due to vestibulo-ocular reflex deficits.6,7 Over the last 10 years, some new entities related to BVH have appeared, such as cerebellar ataxia, neuropathy, and bilateral vestibulopathy syndrome.8 In this case report, a rare clinical case of a simultaneous occurrence of BVH with pressure and sound-induced nystagmus is presented.

Based on the unstimulated labyrinths in the caloric and rotational tests, BVH could be confirmed. There was a clear and definite symptom of a fistula of the right labyrinth. Fistula of the right lateral semicircular canal could not be confirmed, because it had not been detected radiologically. The absence of a lowered threshold of the affected side, during the vestibular evoked myogenic potentials tests, without radiological confirmation, excluded the presence of a fistula in the superior semicircular canal, regardless of the real Tullio phenomenon.9,10 The clinical presentation and laboratory findings can only be explained by the existence of vestibular atelectasis (VA), a collapsed endolymphatic membrane in the area of the ampoules and utriculus, a phenomenon described in 1988, based on an analysis of pathohistological preparations of the temporal bones.11 Before this announcement, Di Nome Aciemo et al. had described a patient with symptoms...
bilateral vestibulopathy and with two types of oscillopsia, one induced by head movement and the other produced by changing the pressure in the right external auditory meatus. Over the last few years, only a few articles on the subject of VA have appeared in medical literature. Although the pathophysiology of VA has not yet been clarified, some authors have presented a very intriguing hypothesis, speculating that a collapse of the perilymphatic membrane in the area of oval window enables a greater adherence of the stapes to the vestibulum, thus strengthening the stimulation of the otolithic senses.

CONCLUSION
The whole concept of VA is not as yet completely and generally accepted. In this case report, we point out an undetectable and etiologically unexplained entity, which, however, allows a diagnostic solution in cases of BVH and a positive fistula symptom without radiological confirmation. Our intention was also to highlight the significance of the vestibular evoked myogenic potentials findings to confirm the VA concept involving the ampoules and the utriculus with a spared sacculus. The diagnosis of VA eliminates the necessity for exploration of the middle ear and detection of any radiologically invisible fistulas.

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BIBLIOGRAPHY