Baro-otolagia Secondary to Cholesteatoma

Revadi Govindaraju; Meenakshi Adaikappan; Raman Rajagopalan

BACKGROUND: Baro-otalgia is a common complaint among passengers in an aircraft, in particular those who had a recent upper respiratory tract infection. The underlying pathophysiology is secondary to unequal aeration of the middle ear cleft with the surrounding atmosphere and it can be explained using Boyle's Law. We describe an unusual presentation of baro-otalgia in a pilot secondary to cholesteatoma obstructing the aditus despite normal middle ear pressure equalization provided by a grommet in the ear.

CASE REPORT: A 26-yr-old pilot with a presenting complaint of conductive hearing loss was diagnosed and treated for congenital cholesteatoma. His hearing improved, but 4 yr later he developed ear pain during the cruising phase of flight at an altitude of 9144 m (30,000 ft) above sea level. This pain persisted until descent to 4876 m (16,000 ft). Despite insertion of a middle ear ventilating tube, he remained symptomatic, requiring further investigation. This led to the diagnosis of recurrent cholesteatoma obstructing the aditus to the mastoid cavity. Upon surgical removal of the cholesteatoma, symptoms resolved.

DISCUSSION: We hypothesize that the recurrent cholesteatoma caused obstruction to normal aeration of the mastoid air cells during the changing atmospheric air pressure, thus producing pain. This is akin to sinus barotrauma instead of the usual pathophysiology underlying barotitis.

KEYWORDS: sinus barotrauma, baro-otitis, congenital cholesteatoma.

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In aviation physiology, one of the environmental changes of physiological significance is the marked change in barometric pressure.[1] In a fixed-wing aircraft, the pilot sets cabin altitude as the aircraft gains altitude to partly compensate for these changes. Most pressurized aircraft are able to maintain a cabin altitude of approximately 2438 m (8000 ft) when the aircraft is flying at an altitude of over 12,192 m (40,000 ft) above sea level. However, this not similar to the barometric pressure at sea level; at 2438 m altitude the barometric pressure is about 565 mmHg compared to 760 mmHg at sea level and thus gives rise to physiological changes of significance.[1] The peak cabin altitude may also vary according to the aircraft model, aircraft class, and distances flown.[2]

The impact of these barometric pressure changes can be described by using Boyle's law. It can affect any enclosed body cavity such as the stomach, intestines, middle ear, and sinuses. As explained by Boyle's law, there is expansion of gas in the middle ear due to the reduction of barometric pressure during ascent of an aircraft. This pressure is equalized with surrounding atmospheric pressure with a normal functioning Eustachian tube (ET) allowing the gas to escape passively every 152 m (500 ft) to 304 m (1000 ft) ascent or when there is a pressure differential of between 15–20 mmHg.[1] On descent, the reverse of Boyle's law occurs. Air in the middle ear contracts, creating a negative pressure. This needs to be equalized by the transfer of more air into the middle ear through the Eustachian tube by the active action of swallowing, yawning, or Valsalva maneuver.

Baro-otalgia is a consequence of the inability to equalize the pressure in the middle ear cleft with the surrounding environment and is a common complaint among passengers, in particular those who had a recent upper respiratory tract infection. We describe an unusual presentation of baro-otalgia in a pilot secondary to cholesteatoma obstructing the aditus to the mastoid cavity despite normal middle ear aeration.

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CASE REPORT

A 26-yr-old civilian pilot presented to the otorhinolaryngology clinic in 2010 with a complaint of reduced hearing in the right ear for the duration of 1 wk. There was no preceding upper respiratory tract infection, history of trauma, or history of ear disease in the past. Otoscopic examination of the right ear revealed a whitish mass behind a normal looking tympanic membrane. The left ear was normal. Tuning fork testing revealed Rinne's positive on the right ear with Weber's test lateralizing to the right, indicating a mild conductive hearing loss. Pure tone audiometry testing revealed a conductive hearing loss with an air-bone gap of 15 dB. High resolution computed tomography (HRCT) scan of the temporal bone showed opacity in the right attic extending to the aditus and mesotympanum. He then underwent surgical exploration via the transcanal endaural approach. Elevation of the tympanomeatal flap revealed cholesteatoma, which was removed and a diagnosis of congenital cholesteatoma was made. There was erosion of the incus noted intraoperatively. Postoperatively he had regular follow-ups and both endoscopic examination and audiometry was normal for the ensuing 4 yr.

He presented again in 2014 complaining of a new symptom, a throbbing right earache while flying. He noted the pain developed only during flights of more than 2 h duration and about 30–45 min after a cruising altitude of 9144 m (30,000 ft) was reached. Pain was absent while at the ground level and during ascent. However, once developed at the cruising altitude, it persisted as the plane made descent until about an altitude of 4876 m (16,000 ft). Though the pain improved thereafter, some discomfort remained for another day. The apparent improvement in symptoms during descent occurred during continuous descent and was not associated with the airplane leveling off for a certain duration of time at the altitude of 4876 m. Otoscopic examination revealed a right dull tympanic membrane with no retraction pocket. Pure tone audiometry was notable for a conductive hearing loss on the right side, but the tympanometry test was normal. A trial of treatment with decongestant nasal drops showed no improvement. This was followed by right myringotomy and grommet insertion, but he remained symptomatic. This gave rise to a high degree of suspicion of recurrence of the congenital cholesteatoma.

A repeat HRCT scan of the temporal bone revealed recurrence of cholesteatoma at the attic extending to the aditus with a normal mastoid air system (**Fig. 1**). Hence the ear was re-explored with elevation of the tympanomeatal flap and the cholesteatoma was removed. Following this procedure he has been symptom free again.

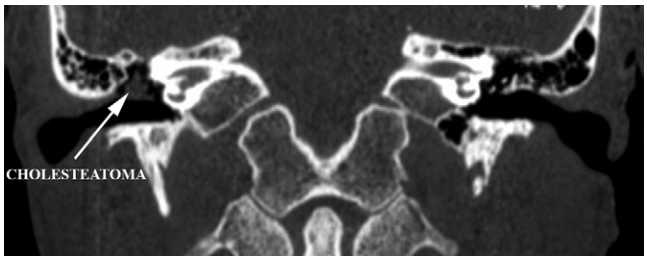

Fig. 1. HRCT of the temporal bone in coronal view with cholesteatoma seen in the epitympanum, but with a clear mesotympanum.

DISCUSSION

In ET dysfunction, negative pressure in the middle ear space can lead to barotitis media, with symptoms typically starting to manifest when the pressure differential is about 60 mmHg.[7] The clinical presentation may vary from ear fullness, pain, hearing loss, tinnitus, vertigo, and nausea with otoscopic findings ranging from mild tympanic membrane retraction and congestion to perforation of the ear drum, which may occur when the pressure differential reaches 100–500 mmHg.[1,7] The severity is believed to be affected by the rate of ascent or descent and also the patient's compensatory mechanism.[1]

The occurrence of baro-otalgia during descent is quite predictable, but it has also been reported to occur as delayed ear pain in a study of aircrews exposed to simulated high altitude.[8] In our case, the timing of the baro-otalgia and the characteristics were not the usual encountered; nevertheless, barotitis needed to be ruled out as it may interfere with the functions of an aircrew.

Despite a normal tympanometry test in the pilot, subclinical ET dysfunction may still exist. A resting middle ear pressure of more than −73 mmHg (−100 mmH_2O) is generally considered pathological ET dysfunction, but studies have shown that barotrauma has even occurred at a resting middle ear pressure below −36 mmHg (−50 mmH_2O).[3] The underlying ET dysfunction is, however, not easily identified using tympanometry alone and, therefore, recent attempts have also been made to use a combination of tubomanometry and the Eustachian Tube Scoring system to assess for subclinical ET dysfunction and its risk in causing barotitis in a group of aviators.[4]

Earache, on the other hand, is not a typical complaint of a patient with a congenital cholesteatoma; rather is it more common for the patient to present with conductive hearing loss and often it is an incidental finding.[5,9,10] This is true as in the case of this pilot during his initial presentation. In some cases of congenital cholesteatoma, acute otitis media and middle ear effusion were the presenting complaint.[10] The manifestation may again differ in case of recurrence of cholesteatoma depending on the type of surgery the patient has undergone, i.e., transcanal, canal wall down, or canal wall up surgery, integrity of the ossicles, location of the disease, and extent of disease. Diagnosis of recurrence in this case was not apparent initially as even myringotomy was normal. It is arguable if second look surgery was indicated in this case to detect early recurrence as the first surgery had complete removal of the disease macroscopically and follow-up otoendoscopy was normal. Current best practice does not favor mandatory second look surgeries for all patients and recommends it if disease removal at prior surgery is uncertain.[6]

In this patient, after treating for suspected barotitis, we would not expect baro-otalgia to persist as the grommet in situ

accommodates the function of ET for both passive and active pressure equalization mechanisms. However, pain did occur and peculiarly started during the cruising altitude and not after descent was begun and improved at a 4876-m (16,000-ft) altitude. The etiology points to the cholesteatoma, but the exact behavior of the cholesteatoma obstructing the aditus is not clear.

We believe his symptoms were akin to sinus barotrauma rather than the typical mechanism in a normal ear. In a paranasal sinus, normal gaseous flow occurs through narrow ostia. As negative pressure builds up in the cavity during descent with no equalization mechanism, as in middle ear and a blocked ostia by intranasal pathology, this leads to sinus pain. This is also known as "squeeze." Sinus pain has also been described during ascent, which can be attributed to expansion of gas within an enclosed cavity with no mechanism for air to enter or escape if the ostia is blocked by intrasinus pathology ("reverse squeeze").[5] The obstructing pathology acts in a ball-valve manner, allowing airflow in only one direction and thus preventing equalization of intrasinus pressure with the ambient atmosphere. Depending on the intrasinus negative pressure, the severity of the symptoms and signs vary and are graded accordingly. As opposed to Teed's classification of middle ear barotrauma, which can be easily diagnosed with otoscopy, the Weismann classification of sinus barotrauma additionally requires imaging to visualize the affected sinus.[7,11,12]

In our patient, we hypothesize that at peak cabin altitude during the cruise phase, the maximally expanded air in the mastoid could not escape secondary to the obstructing cholesteatoma. This caused pain, as seen in a "reverse squeeze." As the gas got absorbed and contracted during descent, a negative pressure developed, causing pain to persist. The obstructing cholesteatoma probably acted as a one-way valve, again preventing further air entering the mastoid cavity to counter this negative pressure build-up, now acting similar to mechanisms seen in "squeeze." This was probably true until increasing barometric pressure during descent finally caused the obstructing cholesteatoma to give way, improving the symptoms. The findings of a normal mastoid cavity without evidence of mucosal thickening or opacity suggest it may not have received the full effect of maximal negative pressure at the ground level, supporting a relief of obstruction and negative pressure build-up somewhere before the ground level was reached. This explanation is taking into consideration changes seen in imaging of aerosinusitis and comparing it to the mastoid cavity.

In conclusion, this phenomenon gave rise to a suspicion for a recurrence of a congenital cholesteatoma. This is important as cholesteatoma left in situ not only destroys the surrounding ossicle, causing conductive hearing loss, but may erode the cochlea and/or the semicircular canals, leading to profound sensorineural hearing loss and vertigo.[9] Other devastating consequences include intracranial complications. Asymptomatic recurrence as in this patient would have been missed until probably later had it not been for the development of this unusual symptom at altitude.

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