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Per-Operative Discovery of Tympanic Paraganglioma: A Case Report

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Abstract

Background: Tympanic paragangliomas are common middle ear benign neoplasias that develop from endocrines cells situated along the great blood vessels of the head and neck, as well as those of the thoracic and lumbar spine. Primary symptoms of tympanic locations include hearing loss and pulsatile tinnitus. Otoscopy often shows a retrotympanic reddish mass, causing tympanic bulging. Computerized tomography scan findings include a tissue density regular mass located at the promontory and limited within the middle ear cavity. Surgical management comprises tumour excision with control of bleeding, with or without prior embolization. **Aim:** The aim of this presentation is to report an unusual case of tympanic paraganglioma. **Case Presentation:** We report the case of a 56-year-old patient whose clinical and paraclinical presentations were consistent with cholesteatoma, though a tympanic paraganglioma was discovered per-operatively. **Conclusion:** In some rare cases, tympanic paraganglioma can be present like a cholesteatoma.

Keywords

Tympanic Paraganglioma, Cholesteatoma, Middle Ear Tumour

1. Introduction

Paraganglia are groups of neural crest-derived structures made up of endocrine cells, dispersed along head and neck blood vessels, as well as thoracic and lumbar spine blood vessels [1]. These tumours grow from paraganglion or glomus

bodies, normal structures of the temporal bone. Glomus bodies originate from nonchromaffin cells; these cells, originating from the primitive neural crest, compose the extra-adrenal neuroendocrine system [2].

A system of classification as to tumour size is described with tympanic, tympanomastoid, infralabyrinthine and intracranial tumours categorized as types A, B, C, and D respectively [3]. This system is helpful when determining the surgical approach to a particular tumor, and for monitoring outcomes and complications.

Paraganglia of the temporal bone are situated on the promontory of the middle ear, along the tympanic branch of the glossopharyngeal nerve, and attached to the adventitia of the dome of the jugular bulb. The frequent symptoms are hearing loss and pulsating tinnitus, most of the time unilateral; adjacent structures are sometimes eroded due to massive vascularization. Tumor growth into the middle ear can destroy the ossicle chain, resulting in conductive hearing loss, facial palsy when fallopian tube is affected, and projection into the outer ear.

Otoscopy often shows a retrotympanic reddish, pulsatile mass that can cause tympanic bulging. Sometimes larger masses may be noted as polyps in the external ear and they may present massive bleeding when handled. Visible mass can be seen in the external auditory canal, originating from the middle ear.

Audiometry can shows conductive hearing loss during early development, but if the cochlea is affected there is a sensorineural hearing loss. During the impedanciometry tumor pulsation will shift the balance-meter needle synchronically to patient's pulse [2].

Tympanic paragangliomas appear on computerized tomography (CT) scan as a tissue density regular mass located at the promontory and limited within the middle ear cavity.

The first treatment is surgical excision, in tympanic tumors, surgery is carried out in the middle ear; an amplified transcanal approach may be enough to entirely remove the tumor with very low-recurrence rates. The various surgical approaches are adapted to the configuration of individual tumors to provide adequate exposure to the limits of the tumor in the mesotympanum, hypotympanum, and mastoid. This surgery approaches with meticulous dissection and hemostasis added with laser and micro-bipolar instruments allows for safe and effective resection of these vascular tumors. In patients where surgery is contraindicated, radiotherapy can be indicated. In cases where the tumor affects aditus and mastoid, a transmastoid approach and removal of the posterior part of the ear canal is indicated.

The clinical presentation and imaging findings of paraganglioma and cholesteatoma can sometimes be similar. This case report presents the case of a 56-year-old patient whose clinical and paraclinical presentations were consistent with cholesteatoma, though a tympanic paraganglioma was discovered per-operatively.

2. Case Report

We received a 56-year-old female patient who presented with chronic left hear-

ing loss for about five years, associated with ipsilateral otorrhoea and intermittently pulsatile high frequency tinnitus. Past medical history was not relevant.

Otoscopic evaluation of the left ear revealed a narrow external auditory canal with whitish debris in the canal. After ear toileting, the tympanic membrane was visible with some inflammatory modifications. The right ear was normal. Rinne's test was negative and the Weber test was lateralized to the left. Pure tone audiometry showed left conductive hearing loss (Figure 1).

Contrast CT scan evaluation of the temporal bones showed a left retrotympanic oval mass, homogeneous without contrast enhancement, with associated scutum erosion and opacification of left mastoid air cells (Figure 2).

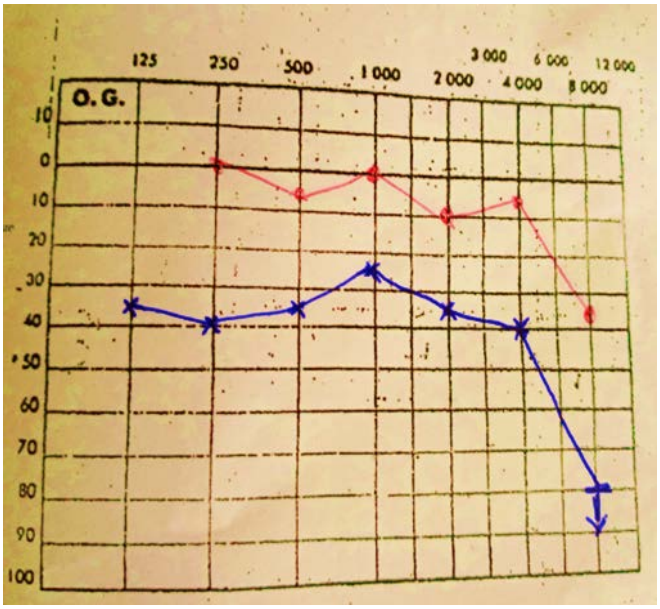

Figure 1. Pure tone audiometry of the left ear: moderate transmissional hearing loss.

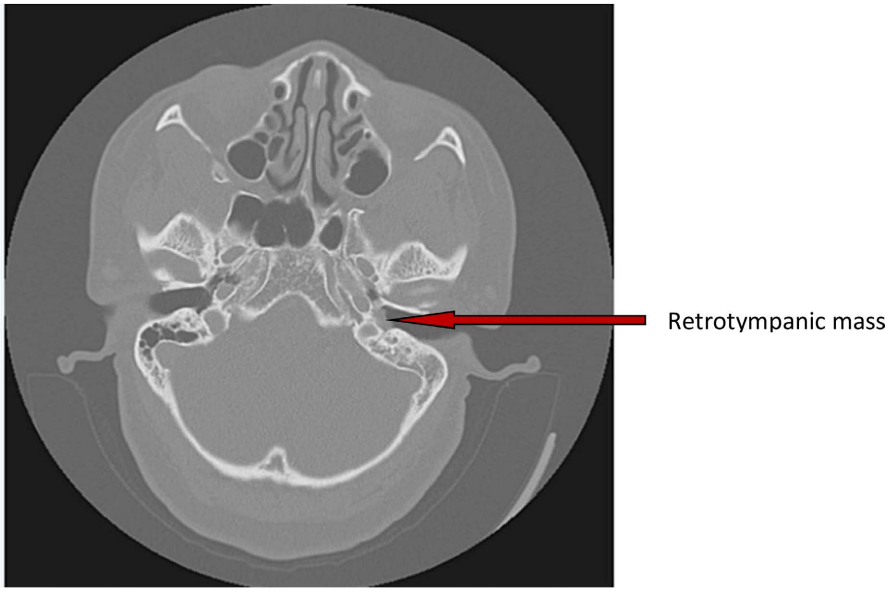

Figure 2. Axial view CT scan of the temporal bones.

The pre-operative diagnosis made was left ear cholesteatoma. A left mastoidectomy was planned and realized. During the procedure, unusual bleeding was noticed, motivating an exploration of the middle ear cavity via a transcanal approach. This exposed a reddish pulsatile mass occupying the entire middle ear cavity, with contact bleeding. A meticulous piecemeal excision of the tumour was done, aided by monopolar cauterization on the microsurgery tools, followed by toileting of the middle ear and mastoidectomy cavities. The procedure was well tolerated, there was no post-operative complication, and the patient was discharged after three days of post-operative care. After 46 days post-operative an otoendoscopic view of the patient tympanic membrane shows a normal tympanic membrane and an empty middle ear cavity (**Figure 3**).

3. Discussion

Tympanic paragangliomas represent the most common middle ear tumours after acoustic neuromas [4]. Macroscopically, paragangliomas are firm, rubbery, nodular or lobulated tumours, reddish and hypervascularized, giving them their pulsatile and haemorrhagic character [5]. This is consistent with the per-operative findings in our patient.

Given the neuroendocrine nature of tympanic paragangliomas, they can be associated with multiple endocrine neoplasia, in which case laboratory studies could reveal abnormalities in 24-hour urine metanephrine, parathormone and calcitonin levels. However, since our working diagnosis was a cholesteatoma, we did not request these tests.

The most remarkable symptoms described in tympanic paragangliomas are pulsatile tinnitus and hearing loss. Interestingly, Falcioni *et al.* [6] reported a case of a patient who presented pulsatile tinnitus, which turned out to be a cholesteatoma. The patient they described reported high frequency, intermittently pulsatile noise, similar to our patient's symptoms. In addition, our patient had intermittent otorrhoea. Otoscopic assessment evidenced whitish debris in the external ear canal, also consistent with cholesteatoma [7].

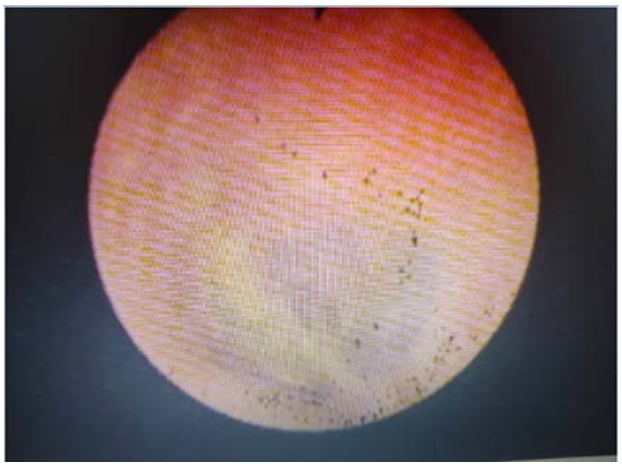

Figure 3. Otoendoscopy of the left ear at 46 days post-operative.

Following ear toileting and suction, we observed an altered tympanum, a finding that could suggest both a cholesteatoma and a paraganglioma. A similar case has been described in Morocco by Rafiq *et al.* [8].

CT scan results for our patient weighed in favour of cholesteatoma. Images showed a retrotympanic mass without contrast enhancement, scutum erosion and opacification of the mastoid air cells. Surgical management consisted of mastoidectomy followed by a transcanal tumour excision, with the aid of monopolar cauterization. However, the use of embolization prior to resection of paragangliomas is recommended [9]. Possible approaches include endaural, transcanal and retroauricular. The extension of the tumour guides the resection process, as it can include drilling of facial bones or the infralabyrinthine bony compartment.

After the annulus is raised and the tympanum displaced, the tumour is usually exposed and gently lifted off the bony walls of the mesotympanum and the ossicles, with the aid of tiny cotton balls. Based on the size of the mass, resection can be done en-bloc or in a piecemeal fashion. The use of fine bipolar cauterization can help to manipulate the tumour and achieve haemostasis. The early post-operative period is usually uneventful. In our case, surgical excision was done using monopolar cauterisation apply on microsurgical tools with piecemeal resection. There was no post-operative complication.

4. Conclusion

Paragangliomas are benign tumours that should be considered in a patient presenting a retrotympanic mass. Clinical and paraclinical presentations are very similar to that of cholesteatoma. In such instances, precautions should be taken prior to surgical excision.

Consent

The informed consent from the patient was obtained by the patient.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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