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Granulation Tissue Mimicking a Glomus Tumor in a Patient with Chronic Middle Ear Infection

ABSTRACT

Objective: To present an unusual cause of pulsatile tinnitus, presenting in a young adult suffering from chronic recurrent foul-smelling discharge from the same ear.

Methods:

Design: Case Report
Setting: Tertiary National University Hospital
Patient: One

Results: A 24-year-old woman presented with pulsatile tinnitus on a background of chronic recurrent foul-smelling discharge. Clinico-radiologic findings seemed consistent with a glomus tympanicum coexisting with chronic suppurative otitis media with cholesteatoma. She underwent tympanomastoidectomy with excision of the mass. Histopathologic evaluation revealed the mass to be granulation tissue.

Conclusion: Pulsatile tinnitus is rarely associated with chronic middle ear infection. Granulation tissue arising at the promontory may mimic glomus tumors when accompanied with this symptom. Despite this revelation, it would still be prudent to prepare for a possible glomus tumor intraoperatively so that profuse bleeding and complications may be avoided.

Keywords: *tinnitus, pulsatile; otitis media, suppurative; glomus tympanicum*

Tinnitus is a complaint experienced by around 10% of the general population. Pulsatile tinnitus, on the other hand, is an uncommon complaint that occurs in 4% of patients experiencing tinnitus.¹ It is a symptom rarely associated with chronic middle ear infection. The aim of this report is to present one such case of pulsatile tinnitus in a patient with chronic recurrent otitis media.

CASE REPORT

A 24-year-old woman presented at the outpatient clinic with a three-year history of continuous unilateral pulsatile tinnitus of the left ear. She also experienced recurrent episodes of otorrhagia associated with ear manipulation from the same ear, as well as recurring foul-smelling ear discharge for six years, with gradually worsening ipsilateral hearing loss. There was also occasional otalgia, headache, and vertigo.

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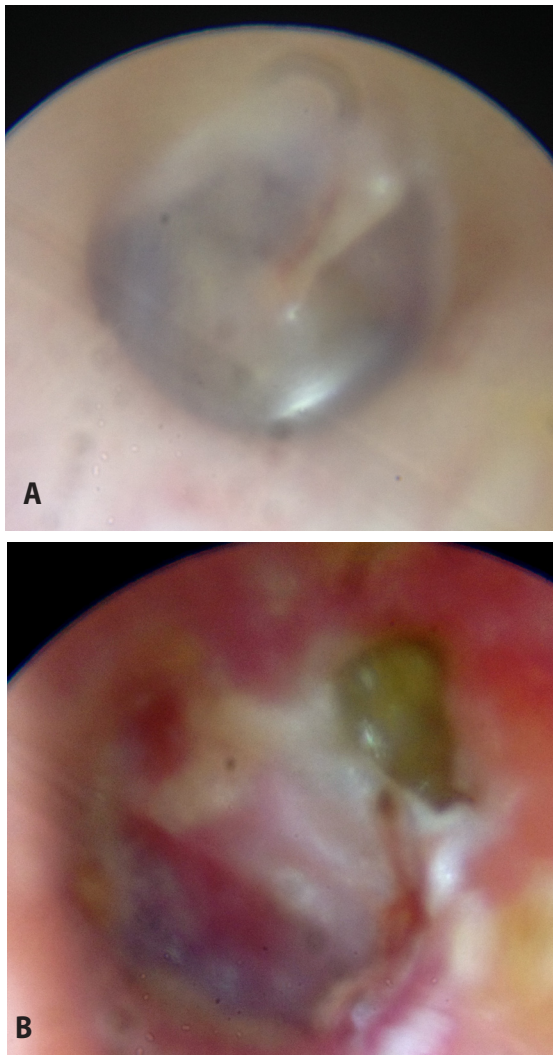


Figure 1. Otoscopic Findings: **A.** Normal otoscopy, right ear. **B.** 20% attic perforation with gross cholesteatoma; remaining tympanic membrane appears thickened, erythematous with violaceous discoloration of the anteroinferior quadrant; ear canal was filled with purulent discharge admixed with whitish debris which was removed.

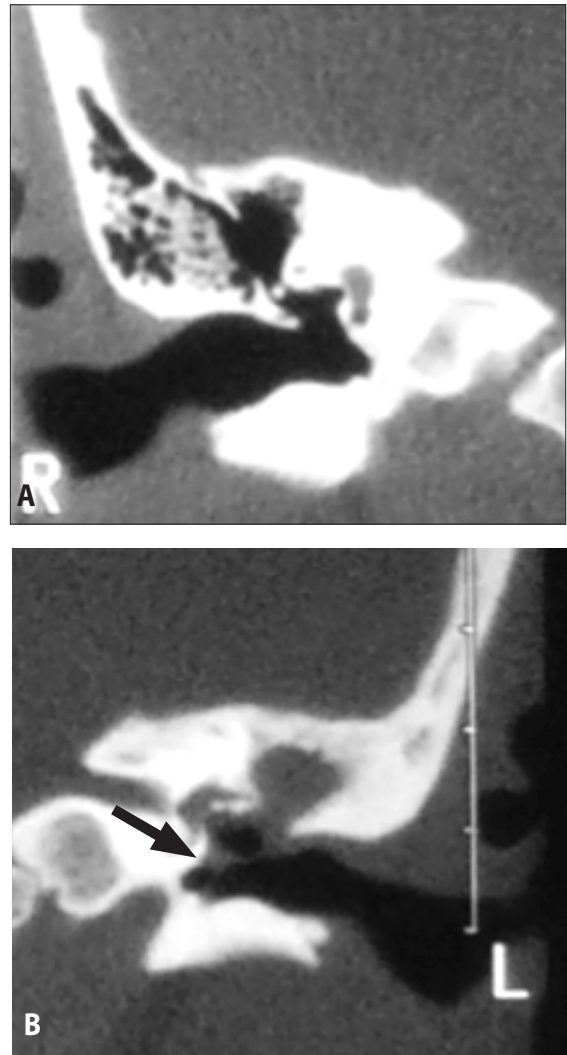


Figure 2. Plain CT scan of the temporal bone. **A.** Coronal cut of the right side showing a sharp scutum with no soft tissue density in the epitympanum. **B.** Coronal cut of the left side showing blunted scutum, erosion of the ossicles, soft tissue density within the epitympanum, and distinct soft tissue mass at the promontory (arrow).

Otoscopy revealed an attic perforation of the tympanic membrane in the left ear with clinical evidence of cholesteatoma. There was also note of a violaceous discoloration of the anteroinferior quadrant of the tympanic membrane. (Figure 1)

Pure tone audiometry and speech testing revealed profound hearing loss on the left ear with no recordable response at maximum stimulus for both air and bone conduction. The right ear had normal hearing acuity.

A non contrast-enhanced Computed Tomography (CT) scan of the temporal bone revealed sclerosed mastoid air cells of the affected side with well-developed air cells on the contralateral mastoid. (Figure 2) The epitympanum of the affected ear was “filled with soft tissue density”

and showed “evidence of an osteolytic process with a blunted scutum and eroded ossicles.” A soft tissue mass was noted in the promontory, distinct from the soft tissue density in the epitympanum. In contrast, the contralateral side showed well-pneumatized mastoid air cells with a sharp scutum and absence of soft tissue densities within the tympanic cavity. The internal carotid arteries on both sides were each separated from their tympanic cavities by the carotid plates. Likewise, both jugular bulbs were separated from the tympanic cavities by their corresponding sigmoid plates.

A Magnetic Resonance Imaging (MRI) with contrast, requested to assess the nature and extent of the mass, revealed a “well-defined, enhancing T1-weighted and T2-weighted imaging-intermediate



subcentimeter focus situated within the left tympanic cavity." (Figure 3) The said mass "did not exhibit restricted diffusion on diffused-weighted imaging. The left mastoid air cells were sclerosed. Petrous pyramids appeared normal and symmetrical. Contrast administration was not followed by an abnormal rise of signal intensity within the nerve, especially its intrameatal portion. There was loss of normal fluid signal in the left lateral semicircular canal" suggesting infiltration.

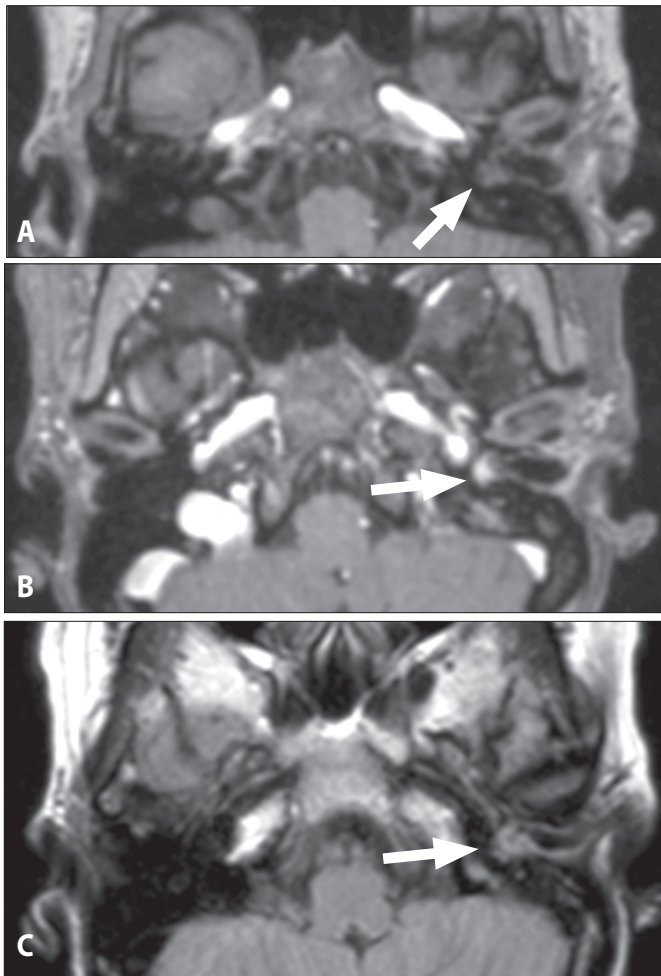


Figure 3. MRI with contrast of the internal auditory canal showing a well-defined mass (white arrow) at the left tympanic cavity measuring 0.9 x 0.6 x 0.7cm (CC x W x AP). **A.** T1-weighted image shows an isointense mass **B.** Gadolinium-enhanced T1-weighted image showing heterogeneous enhancement **C.** T2-weighted imaging showing intermediate enhancement.

Based on these clinico-radiologic findings, our patient was assessed to have a glomus tympanicum and chronic suppurative otitis media with cholesteatoma of the left ear.

A review of systems revealed that she never had any episode of palpitation, abnormal bowel movement, polyuria, polydipsia, weight loss, or blood pressure spikes. To definitively rule out the possibility of a catecholamine-secreting paraganglioma, a catecholamine screen was performed which turned out normal.

She underwent canal wall down mastoidectomy, excision of the middle ear mass, and meatoplasty. Intraoperatively, there was note of a perforation in the attic retraction. A cholesteatoma was seen involving the epitympanum extending into the aditus ad antrum and sinus tympani. Bony destruction was seen at the epitympanum with the ossicles missing; and at the sinus tympani where the stapes footplate was replaced by a cholesteatomatous material over the oval window. There was no evidence of cholesteatoma entering the oval window. The tympanic segment of the facial nerve was dehiscent. The tegmen tympani was intact and the sigmoid sinus was protected by a thick layer of sclerosed bone. The mastoid tip was uninvolved as its air cells were not developed. The middle ear mucosa appeared normal except for the area surrounding the cholesteatoma.

A polypoid, weakly pulsatile mass was seen at the area of the promontory, adjacent to Jacobson's nerve. (Figure 4) The mass bled slowly and continuously but decreased in size and in amount of

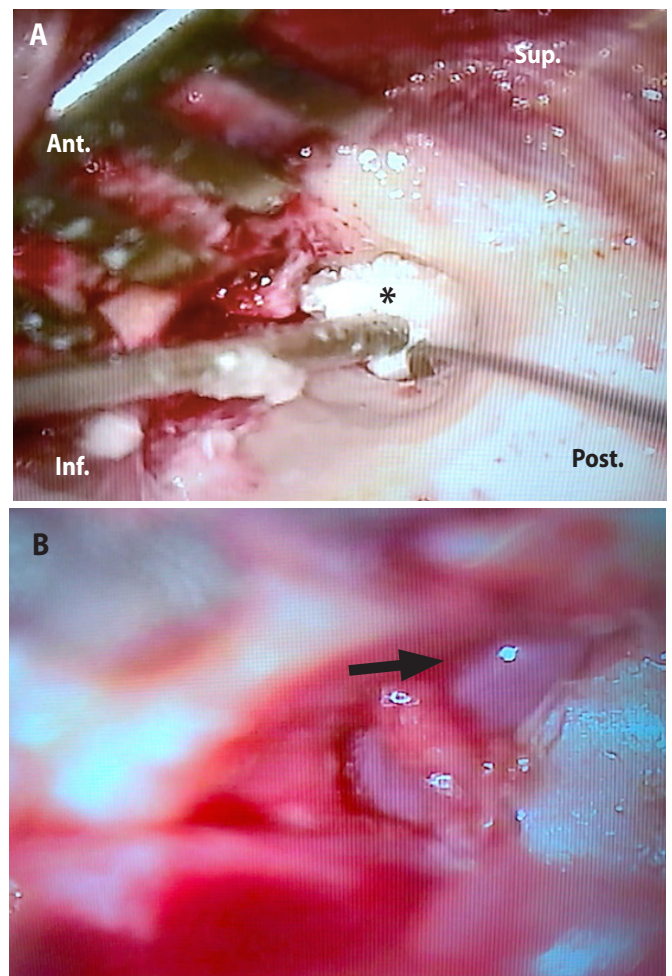


Figure 4. Intraoperative findings: **A.** Removal of the cholesteatoma (asterisk) from the epitympanum extending to the aditus ad antrum. **B.** Pulsating mass found at the cochlear promontory (arrow).

bleeding with application of epinephrine. There was a stalk attaching the mass to the promontory. The mass was excised with cupped forceps and application of epinephrine-soaked cotton was sufficient to control bleeding.

Histopathologic examination showed granulation tissue formation with inflammatory cells composed primarily of plasma cells along with lymphocytes and macrophages. (Figure 5) Immunohistochemistry for kappa and lambda chains were both positive, indicating a polyclonal, non-neoplastic plasma cell population. (Figure 6)

Resolution of pulsatile tinnitus was noted in the immediate post-operative period. No symptom recurrences were noted until 3 months after surgery after which, the patient was lost to follow up.

DISCUSSION

When pulsatile tinnitus accompanies the otoscopic finding of a retrotympenic mass, three entities are considered—arterial anomalies such as an aberrant internal carotid artery, an exposed jugular bulb, or an intratympanic tumor, the most frequent being a glomus tumor.²⁻⁵ Radiological investigation can distinguish these three entities.

Glomus tumors, also known as paragangliomas, are blood-vessel-rich tumors that arise from paraganglion cells of neuroectodermal origin often located near nerves and vessels. Glomus tympanicum is the term used to describe paragangliomas originating from the middle ear, often found on the cochlear promontory along the inferior tympanic nerve, or Jacobson’s nerve. The pulsations generated by the tumor are transmitted to the otic capsule, resulting in increased bone conduction and allowing the sound to be perceived more intensely. Microscopically, a paraganglioma is typically composed of well-formed cuboidal cell nests, or “Zellballen.” (Figure 7) Interspersed between these nests are highly vascularized, fibrous septa. Each cell has an abundant, granular and basophilic cytoplasm. None of these features were seen in the lesion.

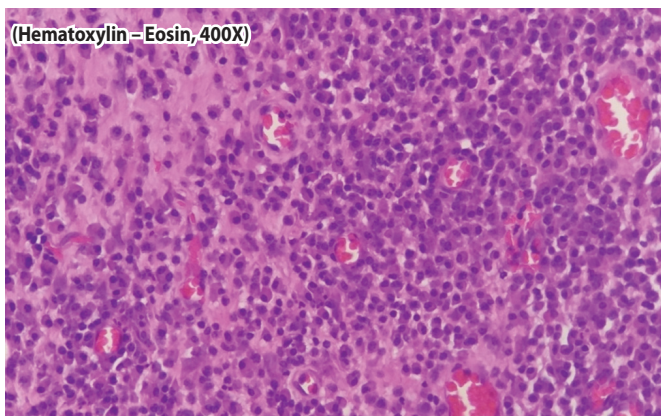


Figure 5. Photomicrographs of the intratympanic mass (Hematoxylin-Eosin, 400X) showing granulation tissue (fibroblasts and new blood vessels) with an inflammatory infiltrate of plasma cells and macrophages.

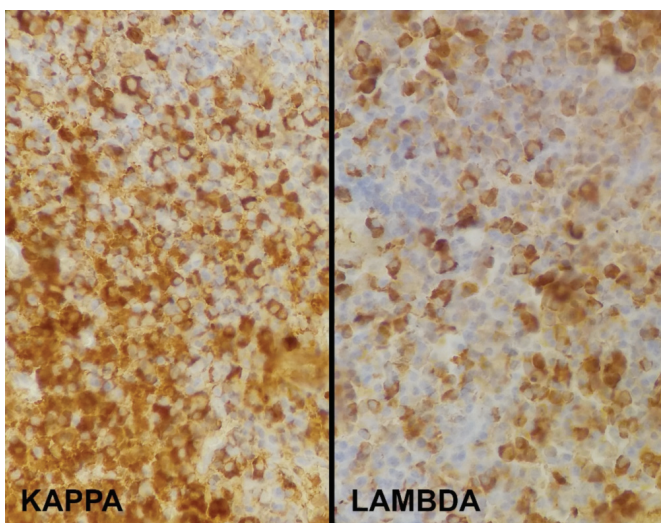


Figure 6. Immunostaining for Kappa and Lambda chains (400X magnification) were both positive.

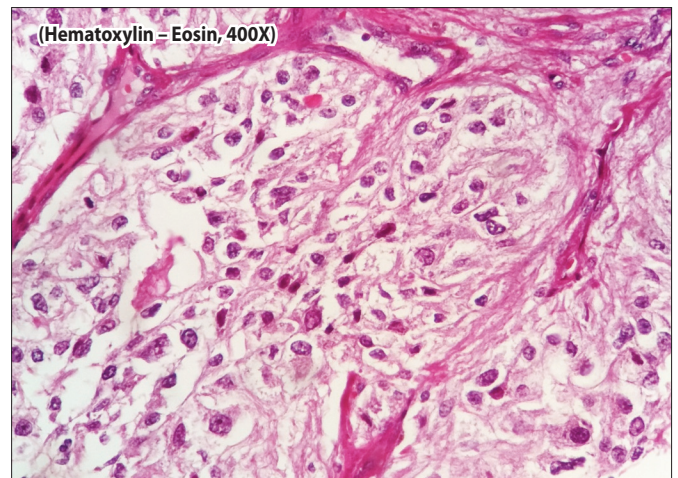


Figure 7. File photomicrograph of a paraganglioma (Hematoxylin-Eosin, 400X) showing typical ‘Zellballen’ formations. (UPCM-PGH Department of Pathology)

An estimated 3-4% of head and neck paragangliomas secrete catecholamines.⁶ Catecholamine-secreting paragangliomas, when undiagnosed, can result in serious perioperative morbidity and mortality. It is therefore important to have a high index of suspicion to reduce complications.

A review of literature associating pulsatile tinnitus with chronic middle ear infection yielded limited results. Falcioni, Taibah, and Rohit in 2004⁷ reported a case of pulsatile tinnitus as a consequence of residual cholesteatoma causing sigmoid sinus compression in a patient who underwent surgery nine years earlier. However, this was not evident



radiographically or intraoperatively in our patient. Several cases of glomus tumors coexisting with chronic otitis media have been reported both in the adult and pediatric population⁸⁻¹⁰ but to our knowledge, pulsatile tinnitus caused by granulation tissue from chronic middle ear infection has not been previously reported.

Granulation tissue is formed as part of the process that follows tissue injury and inflammation, such as is seen in chronic otitis media and cholesteatoma. More frequently associated symptoms of granulation tissue in the middle ear include recurrent foul-smelling ear discharge and hearing loss. Tinnitus, when present, is usually non-pulsatile. Histologically, it is characterized by the proliferation of fibroblasts, angiogenesis or the formation of new thin-walled capillaries, loose extracellular matrix and inflammatory cells such as macrophages.¹¹ According to Michaels, "it is usually particularly prominent in the middle ear under the mucosa covering the promontory from which it frequently protrudes into the external canal through a perforation of the tympanic membrane, forming an aural polyp."¹² The angiogenesis occurring within the tumor could have caused pulsations that transmitted to the otic capsule resulting in pulsatile tinnitus similar to, but with less intensity than a glomus tumor would have caused.

A glomus tumor would present with homogenous and intense enhancement following administration of contrast material owing to its high vascularity. Particularly, a glomus tympanicum should manifest as a small discrete mass arising from the cochlear promontory, and confined to the tympanic cavity.¹³ Granulation tissue on computed tomography should present as a non-expansile soft tissue attenuation material.¹⁴ Some enhancement may also be expected, but not as intensely as a glomus would. However, non-contrast enhanced imaging such as seen in our patient would only show non-specific soft tissue density for either of these two entities causing the confusion in diagnosis.

Cholesteatoma, like granulation tissue, should present a soft tissue density on CT, but unlike granulation tissue, is associated with bony erosion of surrounding structures and a blunted scutum similar to what was seen in our patient.

As in the case of CT, a glomus tumor should also show intense enhancement following contrast administration on MRI. Characteristic of all paraganglioma would be multiple serpentine and punctate areas of signal void variably distributed throughout the mass giving it the so-called "salt and pepper appearance."^{13,15} On the other hand, granulation tissue should present with an intermediate signal on T1-weighted imaging, and a hyperintense T2 signal with diffuse enhancement on delayed post-contrast enhancement.¹⁴ Table 1 summarizes the differences in MRI findings between these two entities as well as cholesteatoma. This suggests that glomus tumors should have similar findings as granulation tissue on MRI.

Table 1. Comparative MRI findings of Glomus, Inflammation and Cholesteatoma

	T1-weighted MRI	T1 – weighted MRI with gadolinium contrast	T2-weighted MRI
Glomus tumor ¹⁴	Hypointensity Areas of signal void ("salt and pepper" appearance)	Hyperintensity Areas of signal void ("salt and pepper" appearance)	Hyperintensity Areas of signal void ("salt and pepper" appearance)
Inflammation/scar tissue ¹⁵	Hypointensity	Hyperintensity	Hyperintensity
Cholesteatoma ¹⁵	Hypointensity	Hypointensity Peripheral rim (matrix)	Hyperintensity

In retrospect, granulation tissue as a cause of pulsatile tinnitus in our case would have been more parsimonious with the history of chronic suppurative otitis media. Nevertheless, a high index of suspicion for a paraganglioma should always be maintained, as the intraoperative complications and profuse bleeding from the latter are much more serious.

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