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CASE REPORT

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Mixed hemangioma of the external auditory canal and the tympanic membrane in a young woman: A case report

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Abstract

Hemangiomas are benign vascular tumors frequently located in the head and neck area, but rarely encountered in the ear, often originally misdiagnosed due to their rarity and absence of specific clinical manifestations. We report the second case of a mixed hemangioma involving the external auditory canal in literature to date.

KEYWORDS

ear, external auditory canal, mixed hemangioma, nose and throat, tympanic membrane

1 | INTRODUCTION

Hemangiomas are the most common benign tumors of vascular origin in the head and neck region. However, their occurrence in the external auditory canal (EAC) and the tympanic membrane (TM) is rare. Hemangiomas are traditionally classified into three major subtypes: (1) cavernous, which are composed of lobules of cystically dilated vascular spaces filled with blood and most regularly occur after the sixth decade of life and are usually linked to previous infection, injury, or hormonal imbalance (2) capillary, characterized by smaller diameter vascular channels that more commonly occur in infancy and progressively regress before the age of 5–6 years and (3) mixed, a combination of both capillary and cavernous.[1,2] Men seem to be more commonly affected than women, while their etiology remains vastly unclear. However, a possible correlation with increased angiogenic peptide basic fibroblast growth factor (bFGF) levels has been suggested.[3]

2 | CASE REPORT

A 38-year-old woman presented to our outpatient clinic complaining of hearing loss and recurrent episode of otorrhea in the right ear for 5 years. The patient did not report otalgia or pulsatile tinnitus. Her medical history was otherwise unremarkable. Otoendoscopy showed a dark purple-not pulsatile-mass partially blocking the EAC, preventing visualization of the tympanic membrane (TM) (Figure 1). Pure-tone audiometry detected moderate conductive hearing loss (Figure 2A). Computed tomography (CT) scan revealed a 6 × 10 mm soft tissue mass in the bone part of the EAC, attached to the TM. There were no signs of bone erosion, middle ear invasion, or presence of exostoses (Figure 3). The patient underwent transcanal surgery under general anesthesia. After resection of the mass, canaloplasty was performed to widen the canal and gain access, followed by myringoplasty to repair the tympanic perforation and skin graft placement to promote rapid tissue healing of the EAC. The excised specimen

was sent for histological analysis, which revealed a mixed hemangioma in the area of the EAC and TM. Microscopic examination of the white-brown polymorphic mass (1.2 × 0.6 × 0.5 cm) showed both capillary and cavernous features, with no evidence of mitotic activity, dysplasia, or malignancy (Figure 4). The postoperative period was uneventful. At 3-month follow-up, the patient showed a significant improvement in auditory testing (Figure 2B). Residual edema of the EAC was present, with good healing of the myringoplasty graft and no signs of disease recurrence.

The present case report conformed to the provisions of the Declaration of Helsinki (as revised in 2013). Written informed consent has been obtained from the patient regarding processing personal information and publication of medical data.

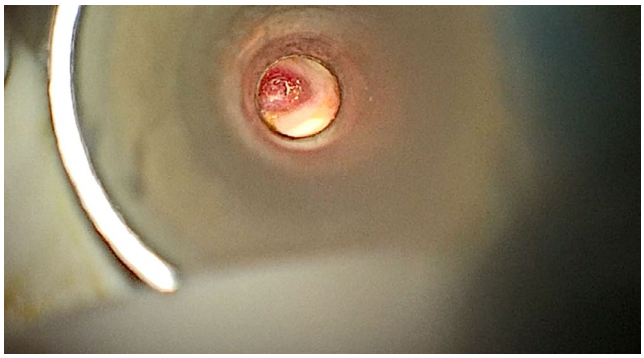

FIGURE 1 Preoperative microscopy image showing a purple non-pulsatile mass obstructing the external auditory canal and obscuring the tympanic membrane

3 | DISCUSSION

Tumoral masses of EAC are unusual in routine ENT clinical practice. The presence of a hemangioma in the EAC was reported for the first time in 1972 by Freedman et al, who described the cases of two men aged 52 and 57 years old, respectively, who presented lesions that initiated from the posterior wall of the EAC and affected the TM.[4] Seven cases of hemangiomas involving the EAC and adjacent TM have been described to date. From a histological perspective, cavernous hemangiomas are the most common (5 out of 7), while capillary hemangiomas are the least frequent, with a single recorded case in literature.[5] Jackson et al. described the only reported case of mixed hemangioma in a 60-year-old woman (Table 1).[6] To our knowledge, this is the second case of mixed hemangioma of the TM and EAC presenting in a young female patient.

Hemangiomas may be asymptomatic, but usually patients complain of hearing loss, tinnitus, aural fullness, and otorrhea. As previous reports have already stated,[1,5,7–9] otoscopy typically reveals a dark purple mass in the EAC; a description consistent with our own finding.

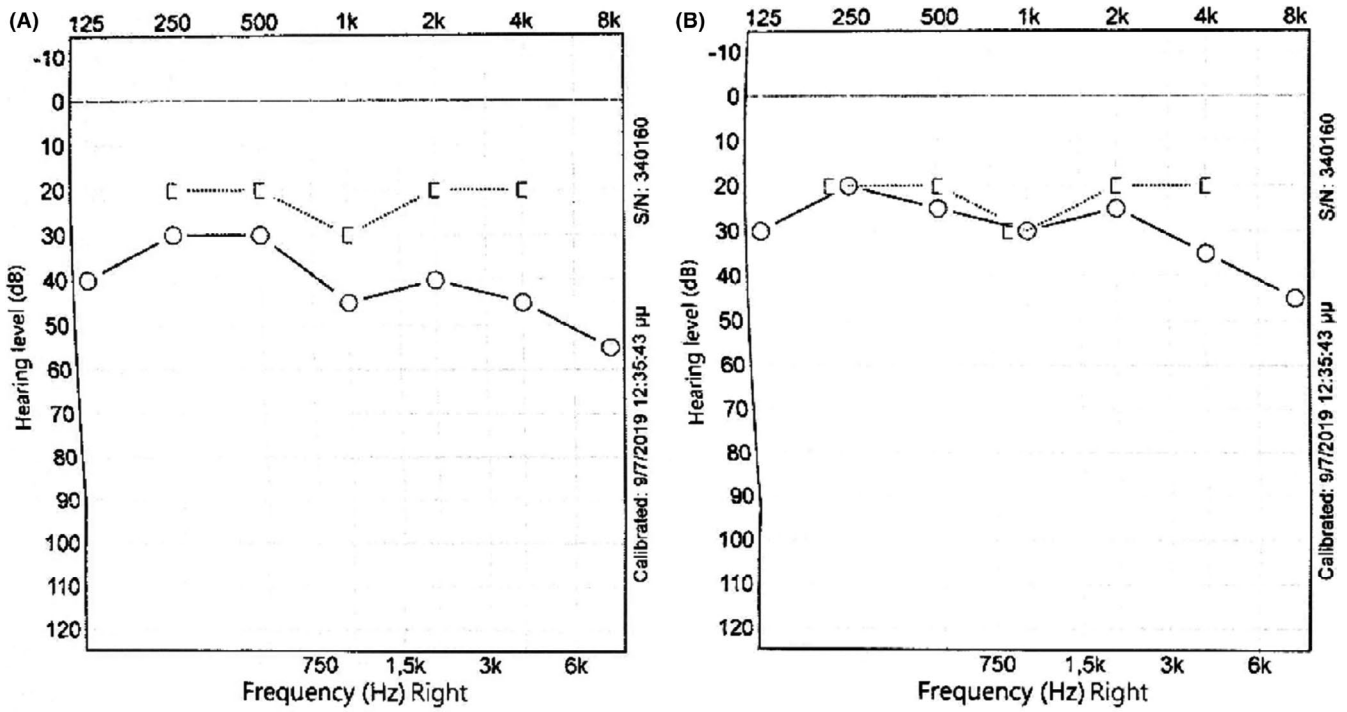

FIGURE 2 (A) Moderate conductive hearing loss in the right ear of the patients was noticed in pure tone audiometry. (B) Following surgery, restoration of hearing was recorded in pure tone audiometry

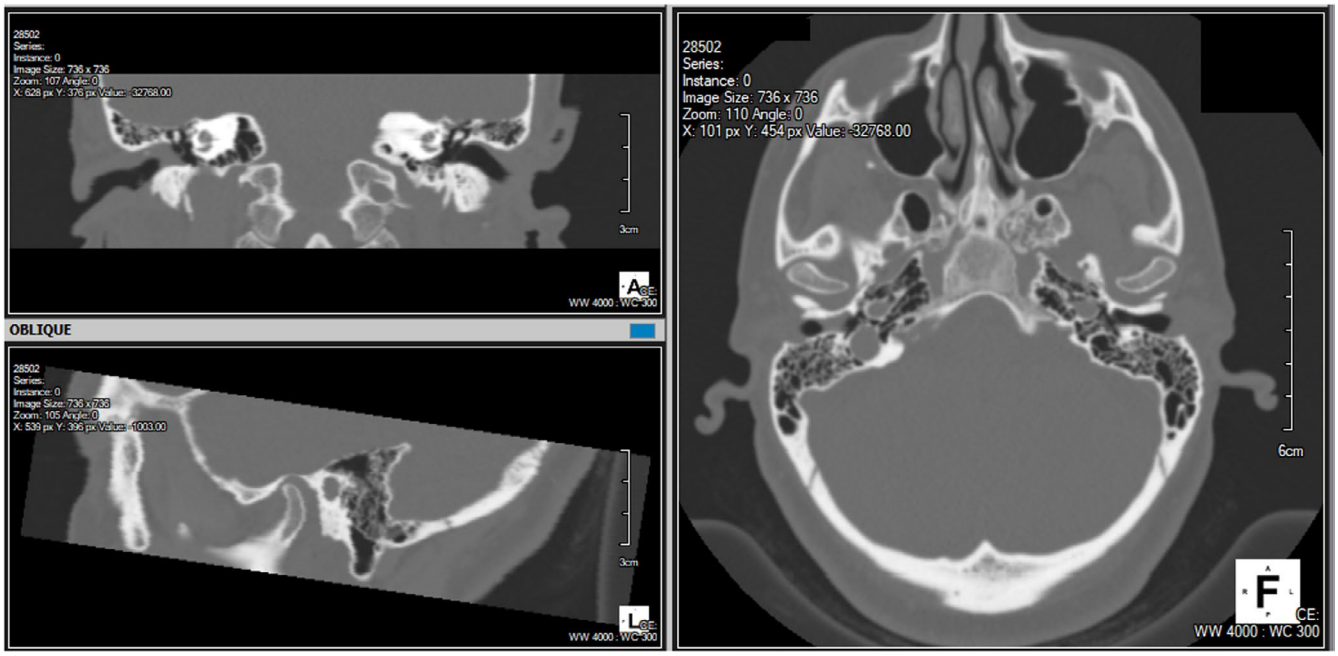

FIGURE 3 Computed tomography imaging revealed a soft tissue mass of the bone part of the external auditory canal in contact with the tympanic membrane causing mild displacement of the membrane

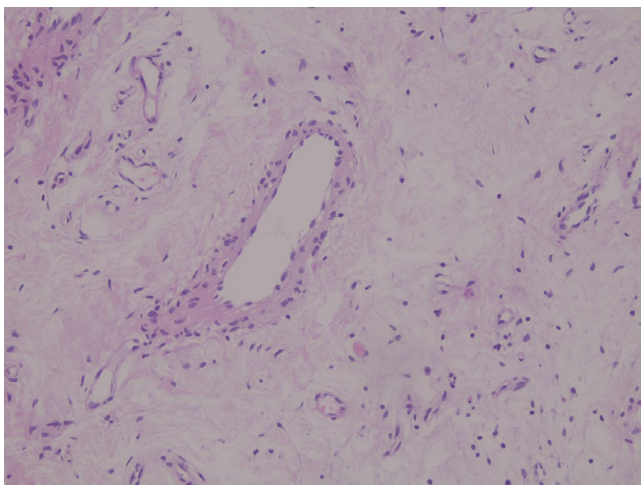

FIGURE 4 Histopathological analysis of the excised specimen in hematoxylin and eosin (H&E) staining (10x/0.25) demonstrated the presence of a large caliber vascular branch surrounded by vascular proliferation mainly by capillary-like blood vessels

A review of the existing literature is summarized in Table 1. In Freedman's case, both patients had normal hearing and the middle ear was unaffected.[4] Kemink et al.[10] also described a case without hearing loss, although the mass extended from the posterior canal up to the TM. In 1990, Jackson et al.[6] described the first case of mixed hemangioma in a patient who had conductive hearing loss and concomitant bone invasion. In this case, the tumor recurred after 2 months and blocked the EAC. Another case by Joshi et al.[7] in 1999 involved a 16-year-old male patient with otorrhea for 2 years with a polypoidal mass, attached to the TM in the left EAC, obscuring its view during examination. Nevertheless, the middle ear was unaffected. In 2007, Magliulo et al.[1] discussed the case of a patient having pulsatile unilateral tinnitus and mixed hearing loss in the right ear. Otoscopy showed a purple, rounded, smooth lesion, affecting the posterosuperior quadrant of the TM and extended to the external ear canal. The mass was soft but not pulsatile, while vestibular examination and the middle ear were normal. The patient was surgically treated, and postoperative audiogram showed recovery of the conductive hearing loss and disappearance of tinnitus. In 2017, Kim et al.[5] described the case of a 54-year-old woman presenting right-sided hearing disturbance and aural fullness; the first case of capillary hemangioma of the EAC and the adjacent TM. Otoendoscopic examination revealed a hard and dark purple mass. Postoperative hearing results showed improvement of the conductive hearing loss.

The differential diagnosis of a hemangioma may be challenging as other vascular lesions of the ear need to be excluded, including arteriovenous malformations, carcinomas, melanomas, pyogenic granulomas, etc.[4,8,9,11] Diagnosis should be based on clinical examination, otoscopy, diagnostic audiological tests, and imaging. In fact, appropriate imaging is vital for proper diagnosis and consequently appropriate treatment. Temporal bone CT performed at high-resolution and small field of view with thin imaging slices (0.5 mm) is regarded as the gold

TABLE 1 Reported cases of hemangiomas involving the external auditory canal and the tympanic membrane of the ear

Author	Age	Sex	Otologic symptoms	Surgical approach	Pathology	Reference
Magliulo, 2007	63	M	HL + T	Excision + Myringoplasty	Cavernous	1
Freedman, 1972	52, 57	M	None	Excision + Myringoplasty	Cavernous	3
Kim SB, 2017	54	F	Fullness + HL	Excision	Capillary	4
Jackson, 1990	60	F	HL	Biopsy + Excision	Mixed	5
Joshi, 1999	16	M	Otorrhea	Excision + Mastoidectomy	Cavernous	6
Kemink, 1983	52	M	None	Excision + Mastoidectomy	Cavernous	9
Lygeros (present case)	32	F	Otorrhea + HL + T	Excision + Myringoplasty	Mixed	

Abbreviations: F, Female; HL, Hearing Loss; M, Male; T, Tinnitus.

standard for the evaluation of bone structure, as well as the location, size, extension, and vascularization of the lesion.[8,9] On the other hand, even though magnetic resonance imaging (MRI) could depict vascular lesions, it does not provide additional details about bone structure, the exact location of the tumor, and the degree of the tissue contained.[1,8]

Treatment of hemangiomas involves surgery, that is, excision and myringoplasty, in order to repair the perforated eardrum. As preoperative embolization provides only relatively low control of the mass, it is not indicated.[1] Following removal of the hemangioma, a skin deficit may arise, making graft use imperative for its restoration. Monitoring of such patients in the form of regular follow-up is considered necessary, as hemangiomas may occasionally recur. When located in the EAC without affecting the TM, a hemangioma causes no symptoms and may remain undiagnosed. Balkany et al,[11] suggested that surveillance of this kind of mass should not be the first choice. The wisest option is early resection in order to prevent the necessity of major TM interventions, potential extension of the lesion, and future recurrence. Their patient, who was initially closely monitored, presented significant increase of the tumor bulk within 18 months and eventually surgery was required for definitive treatment.

In conclusion, hemangiomas of EAC and TM are uncommon and often difficult to differentiate from other malformations. Histological analysis is necessary to confirm the diagnosis. Our case highlights that a thorough clinical and laboratory approach, including audiology testing and high-resolution CT imaging in patients with longstanding ear symptoms, can provide timely diagnosis and management of this rare clinical entity. Complete excision is usually achievable and curative, alleviating patient symptomatology as well as eliminating the probability of recurrence.

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None.

CONFLICT OF INTEREST

The authors have no conflicts of interest to declare.

AUTHOR CONTRIBUTIONS

All authors contributed to the design of this manuscript. MA and AD wrote the first draft. SL and GD edited and reviewed the final manuscript. KG and VD scientifically reviewed the article.

CONSENT

The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient.

DATA AVAILABILITY STATEMENT

Data available on request from the authors.

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