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CASE REPORT

Cavernous Hemangioma of the External Canal, Tympanic Membrane, and Middle Ear Cleft: A Case Report

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Abstract Cavernous hemangioma involving the external canal, tympanic membrane, and middle ear cavity is extremely rare. We present a case of a 45-year-old woman who had progressive right sided decreased hearing, pulsatile tinnitus, and aural fullness of 7 months duration. Microscopic examination, imaging studies, surgical treatment, and histological evaluation are reported. To the best of our knowledge, this is the first case of cavernous hemangioma with simultaneous involvement of the external ear, tympanic membrane, middle ear, and attic reported in English literature.

Keywords Cavernous hemangioma · Vascular lesion · External ear · Middle ear

Introduction

Hemangiomas are common benign vascular tumors of the head and neck area; however they are uncommon in the external ear canal, and rarely involve the tympanic membrane and middle ear [1]. Hemangiomas have been classified into capillary type which is characterized by a lobular unencapsulated proliferation of capillary sized blood vessels, and cavernous type which composed of larger vascular spaces or sinusoids lined by endothelium [2].

Capillary hemangiomas occur more frequently in childhood and cavernous hemangiomas are more common in adults after the sixth decade.

The etiology of hemangiomas is uncertain. In children there may be a genetic predisposition [3]. The growth pattern in children has a proliferative phase in the first year of life followed by spontaneous involution thereafter, in adults this pattern is not well understood [4].

Herein, we report what we believe is the first case of cavernous hemangioma involving the attic, middle ear cavity, tympanic membrane and external auditory canal.

Case Presentation

A 45-year-old woman presented to the otolaryngology clinic with progressive right sided hearing loss, pulsatile tinnitus, and aural fullness of 7 months duration. Microscopic examination of the right ear revealed a reddish sessile mass in the medial portion of the posterior external ear canal pushing the posterior part of the tympanic membrane laterally (Fig. 1a). The lesion was non-pulsatile, and non-blanching with pneumatic otoscopy. Rinne test was negative on the right side and Weber lateralized to the affected ear. Pure tone audiometry showed right sided conductive hearing loss with an average air-bone gap of 30 dB. Facial nerve was intact, other head and neck examination was unremarkable.

Computed tomography (CT) of the temporal bone, showed a 1.2 cm soft-tissue mass involving medial portion of the external canal, posterior part of the tympanic membrane, and middle ear cavity reaching the attic and laying over the second genu of the facial nerve with no

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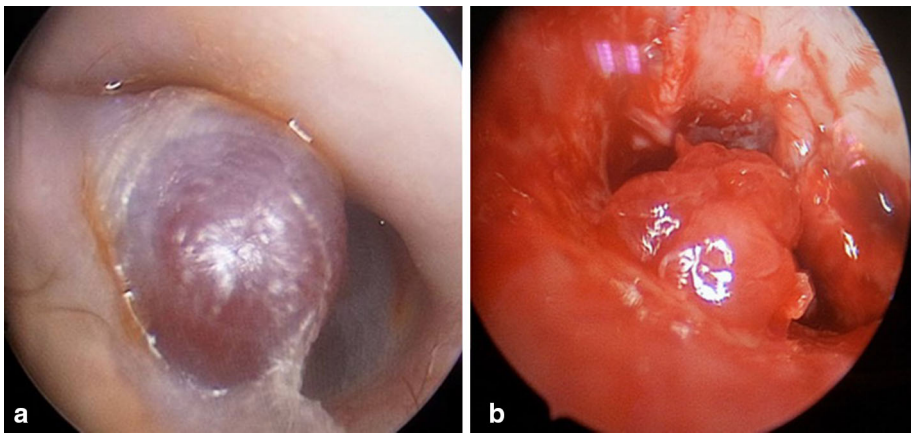

Fig. 1 **a** Endoscopic view of right ear shows a *red-colored*, mass occupying the medial portion of the external canal and pushing posterior part of the tympanic membrane. **b** Endoscopic view of right ear after elevation of tympanomeatal flap shows the vascular mass resting over posterior wall of the external canal and filling the middle ear cavity

evidence of bone erosion (Fig. 2a). The lesion was hyperintense on T2-magnetic resonance imaging (MRI), isointense on T1, and enhanced after gadolinium injection (Fig. 2b).

The mass was excised through canal wall up mastoidectomy. After elevation of the tympanomeatal flap (Fig. 1b), the external canal and tympanic membrane portion of the mass was carefully removed. The middle ear and attic portion was revealed following mastoidectomy with facial recess approach. The mass was filling the attic, pushing the chorda tympani, insinuating between the ossicles and setting over the fallopian canal of the facial nerve just around the second genu. The tumor was completely excised with preservation of the chorda tympani and the ossicles.

Histologically, the mass was non-encapsulated and composed of large cystically dilated, thin walled vascular spaces of variable diameter, filled with red blood cells and lined by flat bland endothelial cells (Fig. 3). Immunohistochemical studies confirmed that the majority of the endothelial lining cells were positive for CD31 immunostain.

The patient made a smooth recovery and was discharged home on the next day. Clinical follow up for 1 year was unremarkable. Her pulsatile tinnitus disappeared, hearing evaluation showed normal air conduction with no air-bone gap and 1 year follow up MRI showed no recurrence.

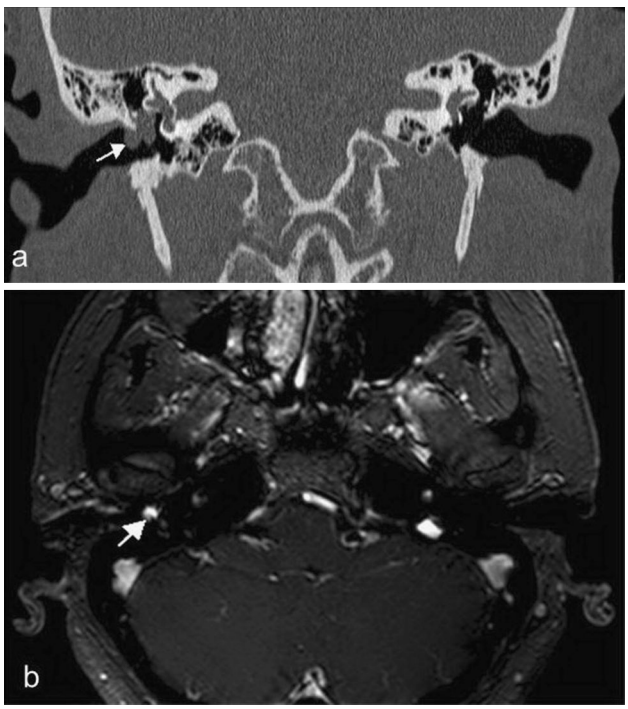

Fig. 2 **a** Computed tomography scan of temporal bone, coronal view shows right side soft tissue mass involving medial part of the external canal, tympanic membrane, middle ear, and reaching the attic (*white arrow*). **b** Magnetic resonance imaging T1 with gadolinium contrast (*axial view*) shows hyperintense lesion in the right ear (*white arrow*)

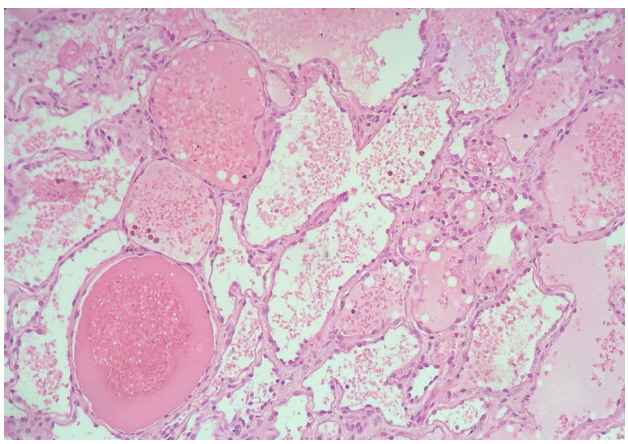

Fig. 3 Histopathologic examination of the specimen showed a cavernous hemangioma composed of dilated, thin walled vascular spaces of variable diameter filled with red blood cells, and lined by flat endothelial cells (H&E, ×200)

Table 1 Reported cases of cavernous hemangiomas involving multiple spaces of the ear

Author	Year	Age (y)/Sex	Location	Surgical treatment
Freedman et al. [5]	1972	52/M	EAC/TM	Endaural excision
Freedman et al. [5]	1972	57/M	EAC/TM	Transcanal excision
Kemink et al. [6]	1983	52/M	EAC/TM	Tympanoplasty, mastoidectomy
Jackson et al. [7]	1990	60/F	EAC/TM/bone	(1) Local excision
				(2) Partial TB resection, STSG
Joshi and Wakode [8]	2000	16/M	EAC + TM	Mastoidectomy
Magliulo et al. [9]	2007	63/M	EAC/TM/malleus	Endaural excision, tympanoplasty
Current case	2014	45/F	EAC/TM/ME/aditus	Tympanoplasty, mastoidectomy

y year, *EAC* external auditory canal, *TM* tympanic membrane, *ME* middle ear, *TB* temporal bone, *STSG* split thickness skin graft

Discussion

Cavernous hemangioma involving the external canal, tympanic membrane, and middle ear cavity is extremely rare. Table 1 summarizes reported cases of cavernous hemangioma involving multiple spaces of the ear [5–9].

To the best of our knowledge, this is the first case of cavernous hemangioma with simultaneous involvement of the external ear, tympanic membrane, and middle ear cleft reported in English literature. Magliulo et al. [9] reported a case of cavernous hemangioma involving the middle ear through a small perforation in the tympanic membrane. The tumor reached only the handle and the short process of the malleus. In our case, the mass extended beyond ossicles to reach the attic and facial nerve canal. There are other solitary case reports of cavernous or capillary hemangiomas involving the middle ear and external ear canal [9–12].

The average age of adult patients with hemangioma of the ear is 55 years and there is a slight male predominance. The external ear canal is the most commonly affected site [13]. Hemangiomas affecting the external canal and tympanic membrane are most commonly the cavernous type and represent around two-third of the reported cases, whereas middle ear hemangiomas, are usually the capillary type which presents 70 % of cases [10, 13].

CT scan is adequate for diagnosis in most of the cases; it shows extension, size, and bone destruction may be assessed [10]. However for lesions with middle ear extension, MRI scan may give more details about the nature of the lesion, extension, relation to the facial nerve and be more helpful for differential diagnosis. Angiogram may be indicated in large lesions when excessive bleeding is expected [9].

Glomus tympanicum, glomus jugulare, attic cholesteatoma, aural polyp, granulation tissue, arteriovenous malformation and malignancy such as carcinoma, should be considered in differential diagnosis of such lesions.

The definitive diagnosis of capillary or cavernous hemangioma is made by histopathologic evaluation [2].

Although close observation is sometimes recommended especially for small asymptomatic lesions, the treatment of choice is complete surgical excision especially in large and symptomatic tumors [3, 12].

Hemangioma may be excised through an end-aural, transcanal, or postauricular approach. In our case; because the mass was insinuated between the ossicles, resting over the facial nerve canal, and reached the attic, canal wall up mastoidectomy was considered to be the appropriate approach gives maximum visualization of the mass to allow complete excision without compromising the facial nerve and ossicles.

Laser excision has been reported to be effective in removing ear hemangioma [14], however this facility may not be available in all centers and needs special technical precautions. We found meticulous dissection, continuous suction, the use of adrenaline soaked cotton pads and early careful identification of the facial nerve and ossicles subsequently used as landmarks are important surgical tips that can help to safely and completely remove hemangioma from the ear.

Close follow up by microscopic examination and/or imaging is recommended to detect early recurrence. Rutherford Leonard [13] and Jackson et al. [7] reported 2 cases of hemangioms that recurred 25 days and 2 months, respectively after the initial resection. We followed our patient for 1 year with no microscopic or radiological evidence of recurrence.

Conclusion

Hemangiomas of the ear are rare benign tumors. We describe the first case of cavernous hemangioma that simultaneously involves the external canal, tympanic membrane, and middle ear cleft. Complete surgical

excision is safe, clinically well tolerated and reduces the possibility of recurrence.

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Compliance with Ethical Standards

Conflict of interest The authors declare that they have no conflict of interest.

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