*Int. Adv. Otol. 2013; 9:(1) 126-130*

CASE REPORT

# Cavernous Hemangioma of the External Auditory Canal: Two Case-Reports and a Literature Review

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**Objective:** To report two unusual occurrence of cavernous hemangiomas of the external auditory canal.

**Study Design:** Case report and review of relevant literature.

**Materials and Methods:** Presentation of patient data, CT and MRI images, surgical treatment and histological findings.

**Results:** Both cases received a treatment regimen of surgical excision. One case involved a relatively large surgical skin defect that was reconstructed with split thickness skin graft, while the other one, with a limited small tumor, underwent excision and was allowed to progress to secondary healing. The postoperative course was uneventful in each case.

**Conclusion:** More than half of hemangiomas occur in the head and neck; however, involvement of the external auditory canal is rare. Surgical resection is the first choice for treatment in symptomatic patients, and close follow-up is recommended to monitor possible recurrence.

*Submitted : 10 October 2011* *Accepted : 21 February 2012*

## Introduction

Hemangioma, also called vascular hamartoma, is the most frequent soft tissue neoplasm in children.[1] About 2% to 10% of hemangiomas develop at birth or in the first few months of life.[2] A unique natural history of hemangioma usually includes a phase of rapid proliferating growth followed by an involution phase to spontaneous regression during the first years of life.[3]

Hemangiomas are commonly classified into the capillary and cavernous subtypes. Capillary hemangiomas disclose tightly packed capillary-like channels, while cavernous hemangiomas are composed of large vascular caliber and are less common. Although more than half of hemangiomas involve the head and neck,[4] and they tend to be located around certain sites that correlate highly with embryologic fusion lines,[5] the involvement of the external auditory canal (EAC) is much less frequently encountered. We present two cases of cavernous hemangioma in the EAC and review the relevant literature.

## Case Reports

### Case 1

A 50-year-old woman complained that her voice had been echoing inside her left ear for 8 months. She initially visited a local clinic, where a mass lesion in the left EAC was found, and she was referred to our outpatient clinic. Otoscopic examination revealed a violet blue mass arising from the inferior bony wall of the EAC (Figure 1A). A hearing test with pure tone

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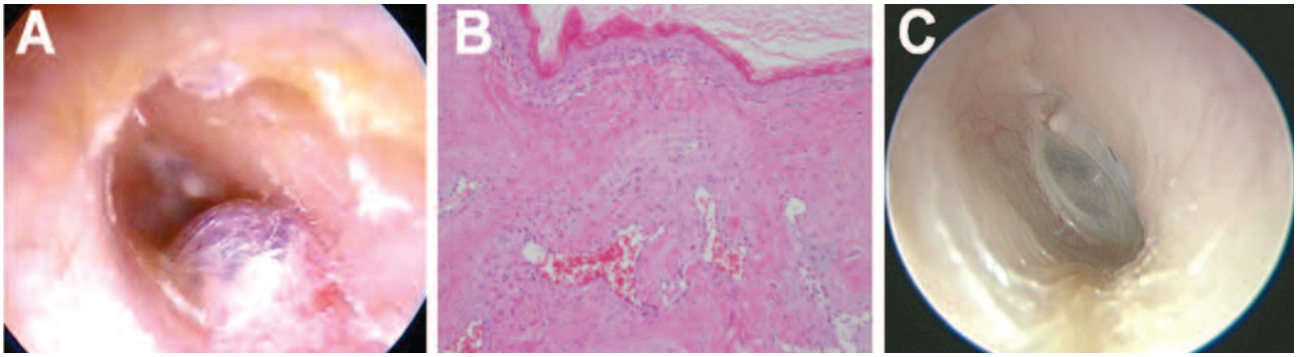


**Figure 1. (A)** A violet mass protruding from the posterior and inferior aspects of the external auditory canal. **(B)** Microphotograph of hemangioma shows characteristic cavernous pattern with numerous wide thin-walled vascular spaces filled with erythrocytes and embedded in the abundant fibrous stroma (H&E stain x200). **(C)** The patient was monitored and disease-free for 24 months with excellent results.

audiometry across all frequencies showed no significant hearing impairment. A high-resolution computed tomography (CT) scan demonstrated a focal mass lesion confined to the bony portion of the EAC, measuring 10 x 4 mm in size (Figure 2A). Subsequent contrast-enhanced magnetic resonance imaging (MRI) disclosed that this localized mass lesion exhibited high vascularity without invasion to the surrounding structures (Figure 2B). The patient denied a past history of chronic diseases or recent traumatic accidents.

Under the impression that this was a benign vascular tumor lesion, a wide excision of the tumor via an endaural approach was performed. By exploring the floor of the bony ear canal, the tumor mass could be well identified and distinguished from the circumferential cutaneous lining in the EAC. Hence, an en bloc excision of the tumor was performed, together with peripheral healthy skin measuring 2 mm in width away from the tumor margin. The skin defect of the exposed bony wall was then covered with a split thickness skin graft. Massive bleeding did not occur, nor was electrical coagulation needed during the operation.

Pathological examination revealed the diagnostic features of cavernous hemangioma by showing proliferation of large tortuous vascular structures varying in size within abundant fibrous stroma (Figure 1B). The wound showed good healing and there was no recurrence at follow-up after 24 months (Figure 1C).

**Case 2**

The second case was an 88-year-old male patient who was referred to our hospital due to one vascular lesion in his right EAC. He was found to have this lesion during a physical checkup at a local clinic, but presented no

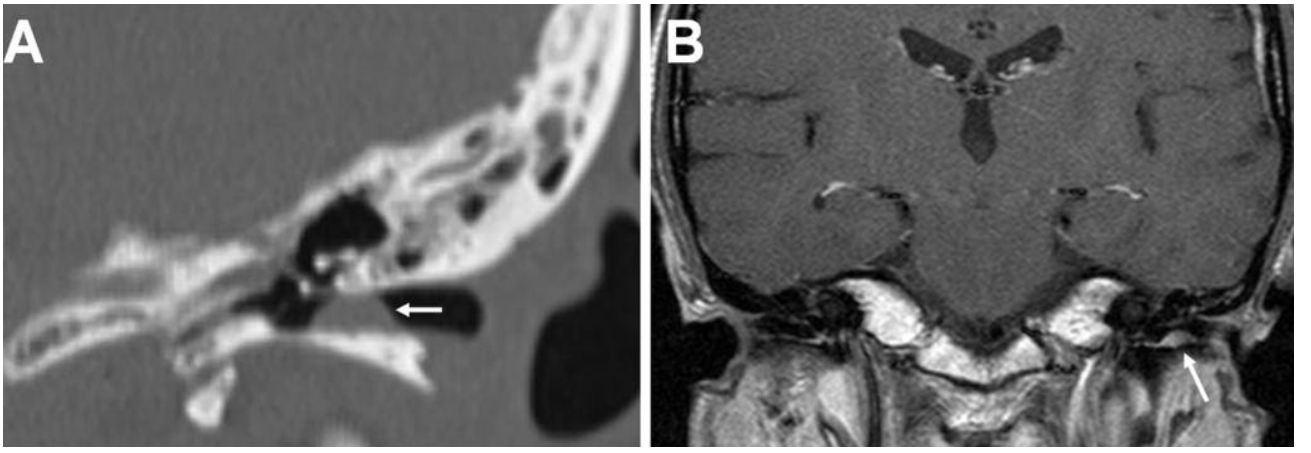


**Figure 2. (A)** Coronal high-resolution computed tomography scan shows that lesion originated from the inferior wall of the bony auditory canal, close to but not invading the tympanic membrane. **(B)** Fat-saturated contrast-enhanced T1-weighted magnetic resonance imaging reveals a solitary hyperintense lesion without an associated soft tissue component and adjacent structure involvement.

specific symptoms. Otoscopic examination of the right ear revealed a solitary, well circumscribed, and vascular tumor situated on the posterior-superior wall of the EAC without contact with the eardrum (Figure 3A).

The involved skin lesion size was small, measuring 4–5 mm in diameter; a simple excision under local anesthesia was performed thereafter. Because the skin defect was of limited size, we packed the wound with an ear wick without further skin or fascia grafting. Pathological examination confirmed the diagnosis of cavernous hemangioma (Figure 3B). That patient had an uneventful postoperative course and remained free of disease at the 12-month follow-up (Figure 3C).

## Discussion

Hemangioma of the EAC is a relatively rare entity. Table I summarizes 22 published cases of hemangioma of the EAC.[1,6-23] In these cases, there is no gender-distributed difference and the age at presentation ranges from 26 to 88 years. Most cavernous hemangiomas grow deeply under the skin with initial normal skin color until later age, which may explain why hemangiomas of the EAC seem to invade those of middle to old age. Pathologically, the cavernous type (14/22; 64%) was more numerous than the capillary type (5/22; 23%), and only one case was presented as a mixed capillary-cavernous lesion (1/22; 5%). In general, most hemangiomas in adults are of the cavernous type, while in infantile hemangiomas, capillary features prevail. Because capillary hemangiomas usually grow in the dermis and are more superficial than cavernous lesions,[24] spontaneous shrinkage over time in childhood may be the reason that capillary hemangiomas of the EAC seldom present in adults. Unlike capillary hemangiomas, cavernous hemangiomas are less likely to spontaneously regress.

Nine cases were shown to involve at least two distinct sites (9/22; 41%); the others were either limited in the tympanic membrane (14%) or confined to the auditory canal (41%). As most of the hemangiomas of the EAC are small in size and usually located inside the ear canal, asymptomatic cases tend to be missed and discovered accidently on x-ray, autopsy, or physical examination, as shown in our second case. The other various symptoms include otalgia, conductive hearing loss, or blood-tinged discharge. Once hemangiomas occur in the tympanic membrane, they will extend laterally to the ear canal rather than medially to the middle ear,[1,15] and they do not have invasive character to destruct the ossicle chain and middle ear as compared with hemangiomas of the tympanic cavity.

Although characteristic vascular findings make it easy to diagnose hemangiomas on inspection, both CT and MR images are believed to be of great value in establishing a differential diagnosis such as carcinoma, glomus tympanicum or jugulare, attic cholesteatoma, aural polyp, granulation tissue, and arteriovenous malformation.[22] The treatment options include surgery, irradiation, laser therapy, cryotherapy, and the instillation of sclerosing agents, but wide surgical excision is still the most effective.[24] Radiotherapy was applied in one huge capillary hemangioma involving the middle and external ear that was unsuitable for surgery.[18] Two cases (2/22; 9.1%) developed recurrence after the initial surgery and pathology: one was mixed capillary-cavernous type[11] and the other was cavernous type.[22] Jackson et al.[11] reported this mixed capillary-cavernous hemangioma, which

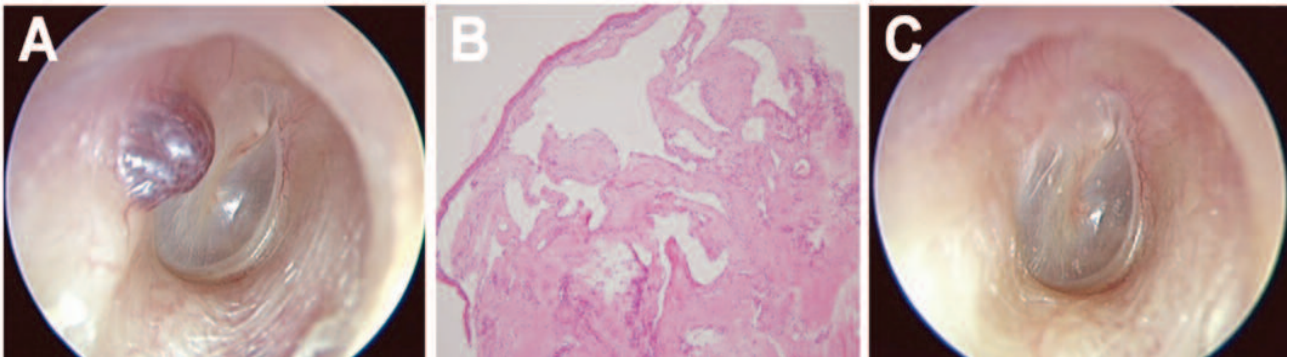


**Figure 3. (A)** Otoscopic examination reveals a lobulated, dark purple mass on the posterior-superior wall of the bony auditory canal. **(B)** The histopathological specimen displays a typical pattern of cavernous hemangioma with dilated vascular channels lined with endothelial cells and surrounded by fibrous tissue (H&E stain x100). **(C)** The patient remains disease-free 12 months after operation.

**Table 1.** Summary of previously reported hemangiomas of the external auditory canal in the literature in English

| References | Year | Age/sex | Location | Pathology | Management | Recurrence |
|---|---|---|---|---|---|---|
| Our case 1 | 2011 | 50/F | EAC | Cavernous | Surgical excision | N (2 years) |
| Our case 2 | 2011 | 88/M | EAC | Cavernous | Surgical excision | N (1 year) |
| 6 | 1972 | 52/M | EAC/TM | Cavernous | Surgical excision | N (5 years) |
| 6 | 1972 | 57/M | EAC/TM | Cavernous | Surgical excision | N (18 months) |
| 7 | 1978 | 63/F | EAC/TM | Capillary | Surgical excision | N ( 2 years) |
| 8 | 1983 | 59/M | TM | Cavernous | Surgical excision | – |
| 9 | 1983 | 52/M | EAC/TM | Cavernous | Surgical excision | – |
| 10 | 1987 | 55/M | EAC | Cavernous | Surgical excision | N (2 weeks) |
| 11 | 1990 | 60/F | EAC/TM/bone | Mixed | Surgical excision | Y (2 months later) |
| 12 | 1997 | 58/F | EAC/TM | Capillary | Observation | No change |
| 13 | 2001 | 78/F | TM | – | Surgical excision | – |
| 1 | 2002 | 67/F | EAC | Cavernous | Surgical excision | – |
| 14 | 2002 | 53/M | EAC | Cavernous | Surgical excision | N (4 months) |
| 15 | 2005 | 51/M | EAC/TM | Capillary | Surgical excision | N (3 years) |
| 16 | 2006 | 72/F | EAC | Cavernous | Surgical excision | N (3 months) |
| 17 | 2007 | 63/M | EAC/TM/MEC | Cavernous | Surgical excision | N (1 year) |
| 18 | 2007 | 26/F | EAC/TM/MEC | Capillary | Radiation | No change (5 years) |
| 19 | 2007 | 31/M | EAC | – | Surgical excision | N |
| 20 | 2008 | 45/F | EAC | Cavernous | Surgical excision | N (3 months) |
| 21 | 2009 | 32/F | EAC | Capillary | Surgical excision | N (4 years) |
| 22 | 2010 | 62/F | EAC | Cavernous | Surgical excision | N (5 months) |
| 23 | 2010 | –/– | EAC/TM | Cavernous | Surgical excision | N (3 years) |

*F = female; M = male;*
*EAC = external auditory canal;*
*TM = tympanic membrane;*
*MEC = middle ear cavity;*
*N = no; Y = yes*

recurred 2 months after the initial local resection with temporal bone invasion; however, there is no association between the histological pattern and its aggressiveness based on the limited number of reported cases and their benign nature originating from the EAC.

Acquired hemangiomas may develop following predisposing risk factors such as radiation and sun exposure.[25-26] Radiation-induced cavernous hemangiomas have been found to increase the risk of hemorrhage when compared to congenital lesions.[26] Our presented cases denied any past history of previous radiation treatment, chemical irritation, and excessive solar exposure.

In summary, hemangiomas arising from the EAC are rare and may be underestimated due to their deep location inside the auditory canal without significant symptoms. Surgical resection is the first choice for treatment in symptomatic patients, and close follow-up is recommended to monitor possible recurrence.

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