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ORIGINAL ARTICLE

A Capillary Hemangioma with External Auditory Canal and Tympanic Membrane Involvement

Fung Wu, Che-Ming Wu, Jen-Liang Su, Kai-Chieh Chan

Division of Otology, Departments of Otolaryngology–Head and Neck Surgery, Chang Gung Memorial Hospital and Chang Gung University, Kweishan, Taoyuan, Taiwan

Hemangiomas of the external auditory canal are extremely rare and the standard management is surgical intervention. Depending on the histopathological characteristics, hemangiomas are classified as either capillary or cavernous types. We present a 36-year-old woman with a capillary hemangioma that involved the external auditory canal and tympanic membrane, with no bony destruction seen on computed tomography. After excision, the patient has remained disease free for 1 year. The relevant literature is reviewed and we summarize the clinical presentation of external auditory canal hemangiomas

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Introduction

Hemangiomas are usually regarded as benign childhood lesions that regress with age. They typically grow in the first year of life and the involution period can range from 6 to 12 years[1]. Hemangiomas are classified as either capillary or cavernous types according to their histopathological characteristics [1]. Capillary hemangiomas consist of closely arranged capillary-like channels, while cavernous hemangiomas are composed of an endothelial lining with large cavernous vascular spaces. They are common on the skin near the parotid glands or in the subglottic area. Hemangiomas in the external auditory canal are exceedingly rare [1]. Here, we report a case of capillary hemangioma involving the external auditory canal and tympanic membrane and review the literature.

Case report

A 36-year-old Taiwanese woman presented with hearing impairment, aural fullness, and otorrhea in the left ear for 1 year. No obvious otalgia was found and the patient denied tinnitus or vertigo. Otoscopy examination revealed that the left external auditory canal was narrow and completely filled by a vascular-like polypoid mass (Figure 1). No facial palsy was observed, and the rest of the otolaryngologic examination was unremarkable. Pure tone audiometry examination revealed a 50-dB threshold of hearing loss with a 25-dB air–bone gap in the left ear. Computed tomography (CT) showed a soft tissue mass located deep in the external auditory canal, adjacent to the tympanic membrane, with no bony destruction (Figure 2). The patient underwent surgical exploration under a microscope. The mass was found to originate from the junction between the annulus of the tympanic membrane and the posterior superior aspect of the bony external auditory canal. It was extirpated completely with excision from its stalk. A myringoplasty was performed to reconstruct the ear drum defect. The specimen measured 1.2 × 1 × 0.8 cm. The histopathological examination showed linear endothelium with closely arranged channels, confirming the diagnosis of capillary hemangioma (Figure 3). The postoperative course was uneventful. No sign of recurrence was observed after 1 year (Figure 4) and the hearing threshold improved to 30 dB without an air–bone gap.

Discussion

Hemangiomas of the external auditory canal and tympanic membrane are rare, and the cause of hemangiomas in the external auditory canal in adults remains obscure. To the best of our knowledge, only 16 adult cases have been published in the English literature (Table 1) [1–15]. Typically, the lesion appears

Corresponding address:
Kai-Chieh Chan, MD
Division of Otology, Department of Otolaryngology, Chang Gung Memorial Hospital and Chang Gung University, No 5, Fu-Shin Street, Kweishan, Taoyuan 333, Taiwan
Phone: 886-3-3281200 Ext.3967; Fax: +886-3-3979361; E-mail: kjchan5109@gmail.com

Approval of the case report was obtained from the Institutional Review Board in Chang Gung Medical Center

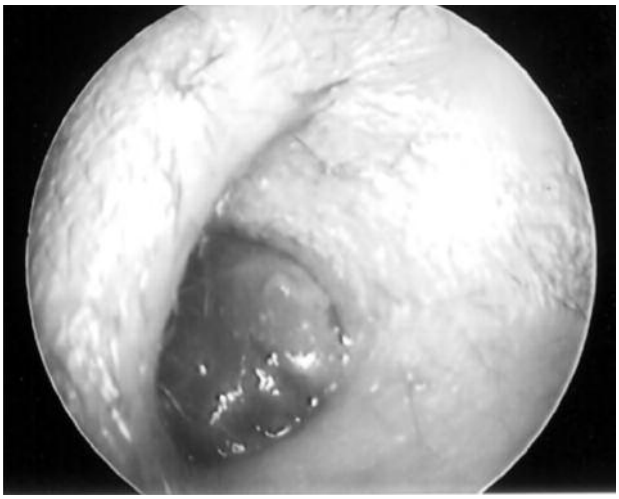

Figure 1. Vascular like polypoid mass occupy the left external auditory canal.

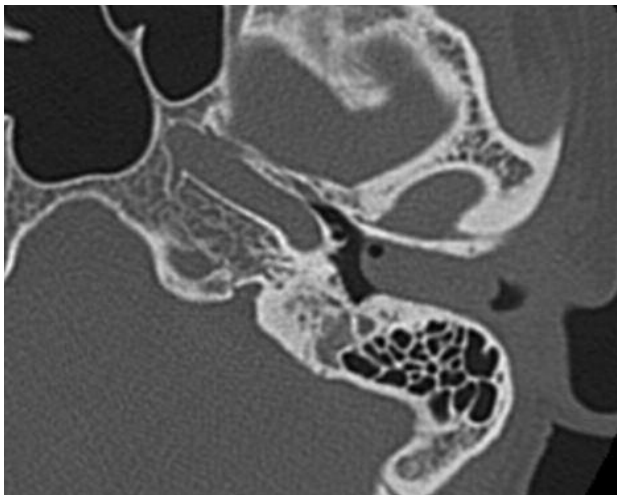

Figure 2. CT shows soft tissue mass locating in the deep external auditory canal and adjacent to the tympanic membrane without bony destruction

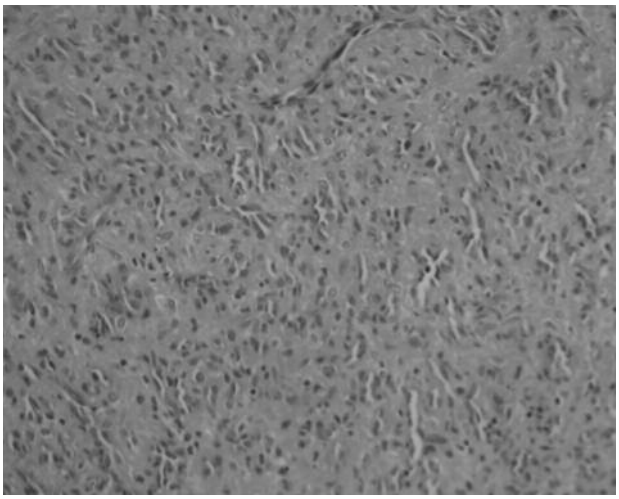

Figure 3. Histopathological examination shows linear endothelium with closely arranged channel, compatible with the diagnosis of capillary hemangioma

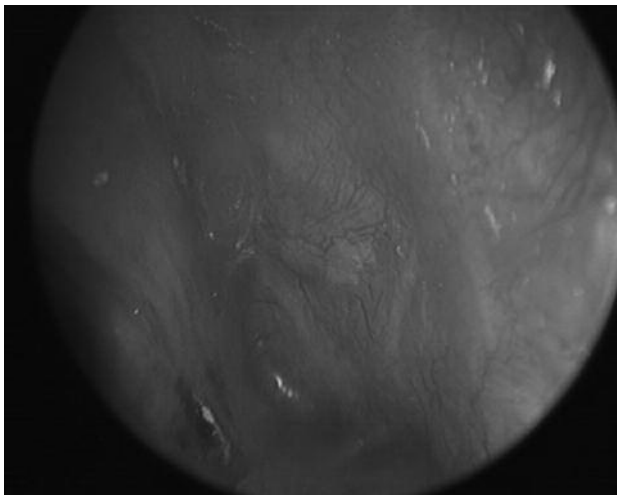

Figure 4. Otoendoscope reveals good epithelization of tympanic membrane and there is no evidence of recurrence sign 1 year postoperatively.

as a red or purple, vascular polypoid mass arising from the posterosuperior portion of the tympanic membrane or external auditory canal wall, although central or anterior origins have also been reported. The most common associated symptoms include hearing loss, otalgia, otorrhea, and ear itching. Including this case, the 17 reported cases consisted of 10 men and 7 women, representing a 10:7 ratio. The ages ranged from 30 to 78 years, although most of the lesions occurred between the sixth and eighth decades of life [1–15]. The tumors were located around the wall of the external auditory canal only in seven patients, involved the tympanic membrane only in four patients, and involved both in six patients. Histologically, the majority of the hemangiomas in the external auditory canal were cavernous hemangiomas. Ten of the 17 cases were cavernous hemangiomas, four were capillary hemangiomas, one was a mixed hemangioma, and two were not classified [1–14].

The previous three capillary hemangiomas were located on either the tympanic membrane or the external auditory canal [3, 10, 15]. Our case is the first capillary hemangioma that involved both the tympanic membrane and external auditory canal. The three cases reported previously underwent surgical excision [3, 10, 15]. The surgical margin was positive in one patient, but no sign of recurrence was observed after 3 years of follow-up [10].

Table 1. The demographic data of the patients with hemangioma in external auditory canal

Author(s)	Age	Sex	Location	Bony erosion	Middle ear involvement	Pathology
Freedman et al. [2]	52	M	EAC, TM	no	no	cavernous
Freedman et al. [2]	57	M	EAC, TM	no	no	cavernous
Balkany et al. [3]	63	F	TM	no	no	capillary
Andrade et al. [4]	59	M	TM	no	no	cavernous
Kemink et al. [5]	52	M	EAC, TM	yes	no	cavernous
Hawke et al. [6]	55	M	EAC	no	no	cavernous
Jackson et al. [7]	60	F	EAC, TM	yes	no	mixed
Bijelic et al. [8]	78	F	TM	no	no	hemangioma
Limb et al. [9]	67	F	EAC	no	no	cavernous
Reeck et al. [1]	53	M	EAC	no	no	cavernous
Hiraumi et al. [10]	51	M	TM	no	no	capillary
Yang et al. [11]	72	M	EAC	no	no	cavernous
Verret [12]	31	M	EAC	no	no	hemangioma
Magliulo et al. [13]	63	M	EAC, TM	no	yes	cavernous
Covelli et al. [14]	45	F	EAC	no	no	cavernous
Murat et al. [15]	30	F	EAC	no	no	capillary

A benign tumor uncommonly invades the nearby bony structures, except via the effect of compression pressure. This also occurs rarely with hemangiomas of the external auditory canal; of the [17] cases, one case of cavernous hemangioma and one case of a mixed-type hemangioma presented with bony erosion [5, 7]. Mastoidectomy and partial temporal bone resection are possible ways of managing a tumor with bony invasion. Middle ear cavity invasion is another possibility. Magliulo et al. reported a patient with a cavernous hemangioma occupying the posterosuperior portion of the tympanic membrane with extension to the osseous external auditory canal [13]. Although the tumor was 3 mm in diameter, tumor invasion into the middle ear cavity was still found at surgery. Middle ear invasion might be correlated more with the location of the tumor, rather than its size.

Although hemangiomas are vascular neoplasms, preoperative angiography is usually unnecessary. In the reported cases, only one patient underwent preoperative angiography, but no vessel for embolization was identified [9]. The patients who did not undergo preoperative angiography still had good intraoperative and postoperative courses, including our patient. The surgical treatment mainly consisted of excision, with tympanoplasty for ear drum involvement. A mastoidectomy was performed in the two cases when bony erosion was suspected in the image examination [5, 7]. The outcomes were mostly excellent and only one case involved recurrence after the first operation [7]. That patient, a 60-year-old woman, was reported by Jackson in 1990; she initially underwent an excisional biopsy for two red, soft-tissue lesions occupying the 4 and 8 o'clock parts of the external auditory canal. A recurrence of the mass was found 2 months later and the external auditory canal was filled. Partial temporal bone resection was performed in the second surgery, as the tumor had infiltrated the bone. The pathologic diagnosis was a mixed capillary- and cavernous-type hemangioma.

In addition to the 17 adult patients described above, Cohen reported an 8-month-old girl who had a left external auditory canal hemangioma in 1972 [16]. Since the presentation and age are very different from the other cases, we discuss this one independently. The infant underwent a biopsy of the left external auditory canal when she was 5 months old and the result suggested a cholesteatoma. At 8 months of age, a red tumor filling the external auditory canal was found. The biopsy showed an inflamed capillary hemangioma, but the hemangioma grew rapidly after the excisional biopsy. After 2 weeks of prednisone therapy, the mass regressed. The patient was treated with steroids for 3 months, but a residual hemangioma over the external canal and middle ear was still noted at 44 months of age. This case suggested a possible medical treatment for external auditory canal hemangiomas, although the tumor did not regress completely.

Conclusion

We present a case of a hemangioma involving both the external auditory canal and tympanic membrane, and review the literature. Although benign courses are found in most cases, bony erosion with the further recurrence is still possible. Surgical management is the definite treatment and in some cases, pre-operative computed tomography evaluation combined with radical procedure is necessitated.

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