Case Report / Olgu Sunumu

A rare vascular tumor of the external auditory canal: the capillary hemangioma

Dış kulak kanalının nadir bir vasküler tümörü: Kapiller hemanjiyom

Hüsamettin Yaşar, M.D.,[1] Haluk Özkul, M.D.,[1] Adnan Somay, M.D.[2]

Department of [1]Otolaryngology, [2]Pathology, Vakıf Gureba Training and Research Hospital, İstanbul, Turkey

A 32-year-old woman presented to our department with a 10-month history of right-sided intermittant otorrhagia. There was no history of hearing loss or pulsatile tinnitus. Otomicroscopic examination revealed a reddish mass arising from the right antero-superior portion of bony canal wall, which measured about 1 cm in diameter. The tympanic membrane seemed to be uninvolved. A computed tomography scan of the temporal bone showed 0.6x0.8 cm diameter soft-tissue mass arising from the right external auditory canal, 0.5 cm away from tympanic membrane. The lesion was excised via a transcanal approach under local anesthesia. The histopathologic assessment indicated a capillary hemangioma. There was no recurrence four years after the surgery. Hemangioma of the external auditory canal is a rare otologic entity. It is commonly classified as capillary or cavernous hemangioma. According to the literature, this case represents the second patient with capillary hemangioma of the external auditory canal.

Key Words: Capillary hemangiomas; computed tomography; tympanic membrane.

Otuz iki yaşında kadın hasta, 10 aydan beri aralıklı olarak sağ kulak kanaması yakınmasıyla kliniğimize başvurdu. İşitme kaybı veya pulsatil çınlama öyküsü yoktu. Kulak mikroskobu ile yapılan muayenesinde, sağ dış kulak kanalı kemik kısmının ön-üst bölümünde kırmızımsı renkte, yaklaşık olarak 1 cm boyutunda, bir kitle saptandı. Timpanik membran tutulumu yoktu. Temporal kemik bilgisayarlı tomografisinde, sağ dış kulak kanalında, kulak zarından 0.5 cm uzaklıkta, 0.6x0.8 cm boyutlarında yumuşak doku kitlesi saptandı. Kitle lokal anestezi altında kanal içi yaklaşımla çıkarıldı. Histopatolojik inceleme sonucu kapiller hemanjiyom olarak bildirildi. Ameliyat sonrası dördüncü yılında hastanın kontrollerinde yineleme yoktu. Dış kulak kanalı hemanjiyomu nadir görülen bir kulak hastalığıdır. Genellikle kapiller veya kavernöz hemanjiyom olarak sınıflandırılmaktadır. Literatüre göre olgumuz dış kulak kanalı kapiller hemanjiyomlu bildirilmiş ikinci olgudur.

Anahtar Sözcükler: Kapiller hemanjiyom; bilgisayarlı tomografi; kulak zarı.

Hemangiomas of the head and neck region have been well-reported.[1-6] and are usually seen as benign childhood lesions that regress with age.[1,7] Hemangiomatous lesions involving the external auditory canal (EAC), on the other hand, are rarely seen.[1-4,6-9] Hemangiomas are commonly classified as either capillary or cavernous.[1,3,5,6] Only thirteen cases of EAC hemangiomas have

Received / *Geliş tarihi:* January 1, 2009 Accepted / *Kabul tarihi:* June 16, 2009

Correspondence / İletişim adresi: Hüsamettin Yaşar, M.D. Eski Londra Asfaltı, Zümrüt Sitesi, A Blok, No: 101, D: 14, 34182, Şirinevler, İstanbul, Turkey.
Tel: +90 212 - 534 69 00 Fax *(Faks):* +90 0212 - 621 75 80 e-mail *(e-posta):* husamettinyasar@yahoo.com

Presented at the 29th Turkish National Congress of Otorhinolaryngology Head and Neck Surgery, May 26-31, 2007, Antalya, Turkey
29. Türk Ulusal Kulak Burun Boğaz ve Baş Boyun Cerrahisi Kongresi'nde sözlü sunum olarak sunulmuştur, 26-31 Mayıs 2007, Antalya, Türkiye.

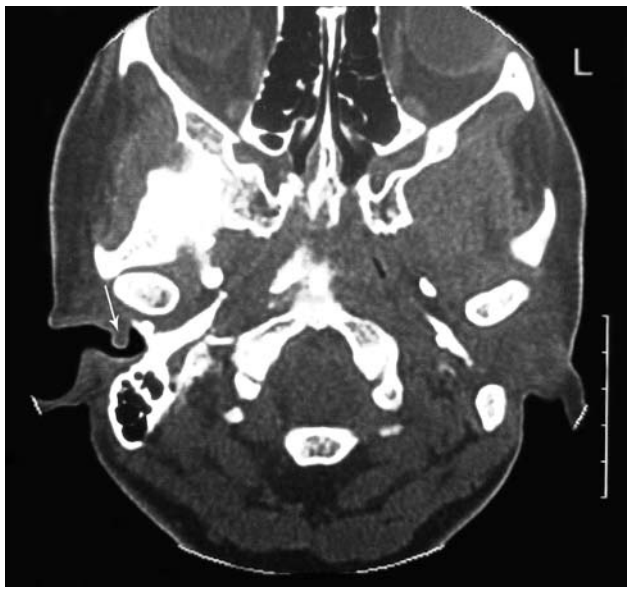

Fig. 1. A computed tomography scan of the temporal bone showing a small soft-tissue mass in the right external auditory canal (arrow).

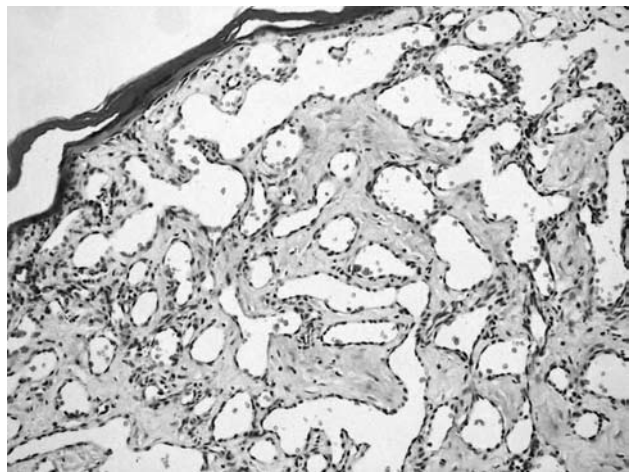

Fig. 2. Photomicrograph showing the typical appearance of a capillary hemangioma (H-E x 200).

been reported up to this date; one of them non-classified, one capillary and one of mixed type.[1-12] A very unusual case is presented here with the relevant literature.

CASE REPORT

A 32-year-old woman presented to our department with a ten-month history of intermittent otorrhagia on the right-side. There was no history of hearing loss or pulsatile tinnitus. The otomicroscopic examination revealed a reddish mass arising from the anterosuperior portion of bony canal wall, which measured about 1 cm in diameter. The tympanic membrane seemed to be uninvolved. Hearing evaluation was also normal. A computed tomography (CT) scan of the temporal bone showed a soft-tissue mass of 0.6x0.8 cm diameter arising from the right EAC, 0.5 cm away from tympanic membrane (Fig. 1). The lesion was excised via a transcanal approach under local anesthesia. Hemostasis was obtained with

Table 1. Reported cases of external auditory canal hemangioma: demographic data and lesion location[1-12]

Author(s)	Age/ gender	Lesion, location	Canal wall involvement	TM involvement
Freedman et al.[10]				
Case 1	52/M	EAC/TM, right	Posterior	SP
Kemink et al.[9]	52/M	EAC/TM, right	Posterior	SP
Hawke and van Nostrand[8]	55/M	EAC, right	Inferior	–
Krueger and Porto[12]	50/M	EAC, left	Superior posterior	–
Jackson et al.[1]	60/F	EAC/TM/Bone, not reported	Inferior anterior and posterior	–
Limb et al.[2]	67/F	EAC, left	Superior posterior	–
Reeck et al.[3]	53/M	EAC, left	Inferior posterior	–
Yang et al.[7]	72/F	EAC/TM, left	Roof of the medial end of the bony canal	–
Magliulo et al.[6]	63/M	EAC/TM/Middle ear, right	Superior posterior	SP
Verret et al.[11]	31/M	EAC, right	Filling the EAC	–
Redaelli de Zinis et al.[4]				
Case 1	74/M	EAC, left	Posterior	–
Case 2	56/M	EAC, left	Inferior anterior	–
Covelli et al.[5]	45/F	EAC, left	Superior anterior	–

M: Male; F: Female; EAC: External auditory canal; TM: Tympanic membrane; SP: Superior posterior; (–): The tympanic membran was not involved.

Table 2. Reported cases of external auditory canal hemangioma: symptoms, treatment, pathology, follow-up/recurrence[1-12]

Author(s)	Symptoms	Treatment	Pathology	Follow-up/ recurrence
Freedman et al.[10]				
Case 1	None	Excision	Cavernous	5 years/(–)
Kemink et al.[9]	Not reported	Tympanoplasty with mastoidectomy	Cavernous	Not reported
Hawke and van Nostrand[8]	Otorrhea, otorrhagia	Transcanal excision	Cavernous	2 weeks/healed
Krueger and Porto[12]	Plugged hearing, local discomfort	Transcanal excision	Benign capillary	Not reported
Jackson et al.[1]	External otitis	Excisional biopsy	Mixed	2 months/ recurrence and temporal bone resection
Limb et al.[2]	Hearing loss, tinnitus, aural fullness	Excision via endaural approach	Cavernous	Not reported
Reeck et al.[3]	Hearing loss, tinnitus	Excision via endaural approach	Cavernous	4 months/(–)
Yang et al.[7]	Incidentally found, hearing loss	Transcanal approach	Cavernous	3 months/(–)
Magliulo et al.[6]	Tinnitus	Resection via endaural approach	Cavernous	1 year/(–)
Verret et al.[11]	Hearing loss, sensation of a foreign body	Resection via postauricular approach	Not reported	Not reported/(–)
Redaelli de Zinis et al.[4]				
Case 1	Otorrhagia, pain	Patient refused surgery	A-V vascular lesion	4 years/(–) changes
Case 2	Otorrhagia, pain	Patient refused surgery	Not reported	10 years/(–) changes
Covelli et al.[5]	Hearing loss, aural fullness	Excision via endaural approach	Cavernous	3 months/(–)

A-V: Arteriovenous; (–): Relapse does not exist.

bipolar cautery. No sutures were needed to close the wound primarily. The tumor was a reddish, soft mass and measured 0.6x0.7x0.8 cm. Histopathologically, the lesion demonstrated a keratinized, stratified, squamous epithelium surfacing the dense fibrous, connective tissue. Within this connective tissue, there was a network of numerous variably dilated capillary vessels. No significant mitotic activity was evident in either the vessels or the stroma. The histopathologic assessment indicated a capillary hemangioma (Fig. 2). The postoperative period was uneventful. There was no recurrence four years after the surgery.

DISCUSSION

Hemangiomas are commonly classified as either capillary or cavernous. Capillary hemangiomas consist of closely arranged capillary-like channels, and may typically occur in the skin and subcutaneous tissues, lips, liver, spleen, or kidneys. Cavernous hemangiomas are composed of large cavernous vascular spaces and often appear in the skin, mucosal surfaces, and internal organs.[3,5,6] Hemangioma of the EAC is a rare otologic entity.[1-4,6-9] There are only 13 reported cases in the literature (Table 1, 2). Freedman et al.[10] first described a case of cavernous hemangioma of the EAC in 1972. A second case

was reported by Kemink et al.[9] in 1983. Most of the reported cases were about cavernous hemangiomas. Only one mixed type and one capillary hemangioma has been reported.[1,12] Although the number of total cases is few, there are similarities among these cases that may indicate characteristic tendencies of this disease entity. External auditory canal hemangiomas seem to occur in individuals in their sixth and seventh decades of life.[1-3,6,7] There is a male preponderance of 2:1.[1,2,6] Most commonly, these masses arise from the posterior canal wall and adhere partially to the tympanic membrane.[1-4,6] Hemangioma may invade both the EAC and the tympanic membrane.[1,6,7,9,10] The presenting symptoms include bloodstained otorrhea, tinnitus, aural fullness, and hearing loss.[1-6,8-12] It may also be asymptomatic and found incidentally.[7] In the reported cases, the otomicroscopic examination revealed a purple, red or violaceous vascular mass in the EAC.[7] The size of the tumors varied from 0.5 to 3 cm in diameter and they partially or completely filled the EAC.[1,7] A high resolution CT scan of the temporal bone is helpful to delineate the extent of the tumor preoperatively. The skull base relationships can be discerned, the osseous erosion can be evaluated, and the differential diagnosis can be narrowed with this imaging modality.[3-8,11,12] In case of an advanced lesion, an angiography may be necessary to identify the blood supply to the mass and to embolize any feeding vessels preoperatively.[7] However, Limb et al.[2] stated that preoperative embolization is not required.[5,6] In our case, although the mass seemed vascular in otomicroscopy, the temporal bone CT showed a soft-tissue mass arising from bony EAC. So we didn't consider angiography as a diagnostic method. The differential diagnosis may include attic cholesteatoma with aural polyp, granulation tissue, intradermal nevus, glomus tympanicum, glomus jugulare, arteriovenous malformation, high jugular bulbus, aberrant internal carotid artery and carcinoma.[3-5,7,10,13] The treatment of choice is a complete excision.[2,3,5-7,12] Hemangiomas of the EAC do not usually have invasive features. The middle ear is generally unaffected by external auditory hemangiomas, although hemangiomas of the internal auditory canal or the middle ear may protrude into the external auditory canal.[2,3] Treatment modality depends on the extent of the tumor. Small lesions confined to the EAC can be treated with a transcanal excision. Large lesions extending to the tympanic membrane and mastoid require a tympanoplasty together with a mastoidectomy and may even necessitate a partial temporal bone resection.[4,7] Recurrence of the EAC hemangioma because of inadequate excision has been reported.[7]

Although hemangiomas arising solely from the EAC are rare entities, they should be considered in the differential diagnosis of vascular masses in the EAC.

REFERENCES

1. Jackson CG, Levine SC, McKennan KX. Recurrent hemangioma of the external auditory canal. Am J Otol 1990;11:117-8.
2. Limb CJ, Mabrie DC, Carey JP, Minor LB. Hemangioma of the external auditory canal. Otolaryngol Head Neck Surg 2002;126:74-5.
3. Reeck JB, Yen TL, Szmit A, Cheung SW. Cavernous hemangioma of the external ear canal. Laryngoscope 2002;112:1750-2.
4. Redaelli de Zinis LO, Galtelli C, Marconi A. Benign vascular lesions involving the external ear canal. Auris Nasus Larynx 2007;34:369-74.
5. Covelli E, De Seta E, Zardo F, De Seta D, Filipo R. Cavernous haemangioma of external ear canal. J Laryngol Otol 2008;122:e19.
6. Magliulo G, Parrotto D, Sardella B, Della Rocca C, Re M. Cavernous hemangioma of the tympanic membrane and external ear canal. Am J Otolaryngol 2007;28:180-3.
7. Yang TH, Chiang YC, Chao PZ, Lee FP. Cavernous hemangioma of the bony external auditory canal. Otolaryngol Head Neck Surg 2006;134:890-1.
8. Hawke M, van Nostrand P. Cavernous hemangioma of the external ear canal. J Otolaryngol 1987;16:40-2.
9. Kemink JL, Graham MD, McClatchey KD. Hemangioma of the external auditory canal. Am J Otol 1983;5:125-6.
10. Freedman SI, Barton S, Goodhill V. Cavernous angiomas of the tympanic membrane. Arch Otolaryngol 1972;96:158-60.
11. Verret DJ, Spencer Cochran C, Defatta RJ, Samy RN. External auditory canal hemangioma: case report. Skull Base 2007;17:141-3.
12. Krueger RA, Porto D. Pathologic quiz case 2. Benign capillary hemangioma. Arch Otolaryngol Head Neck Surg 1988;114:1480-3.
13. Cağici CA, Yilmaz I, Ozlüoğlu L, Kayaselçuk F. Intradermal nevus of the external auditory canal: a case report. Kulak Burun Bogaz Ihtis Derg 2004;12:91-4.