

Hemangiomas of the external auditory canal: a literature review and two new case reports

Case Report

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Abstract: Aim: The aim of this paper is to present two case reports of patients with hemangiomas of the external auditory canal, and to overview all cases published in English language literature so far. Methods: Two patients with hemangiomas of the external auditory canal have undergone clinical and morphofunctional examination, surgical procedures, histopathological examination, and postoperative follow up. Results: In a 38-year-old female patient transmeatal approach was used to remove the cavernous hemangioma of the external auditory canal. The postoperative period was eventless, with normal morphofunctional findings during a 5-month postoperative follow-up period. In the second patient, a 68-year-old female, a capillary-cavernous hemangioma was removed using retroauricular approach. Conclusion: Cavernous hemangiomas are rare lesions of the external auditory canal. In the morphological diagnosis, computed tomography of the temporal bones is the leading diagnostic procedure, along with otoendoscopy and endocranial magnetic resonance imaging which are important in evaluation of the spread of the lesion. Biopsy of vascular lesions is not recommended. Complete surgical excision of hemangioma of the external auditory canal is the therapy of choice with a minor risk of hearing impairment.

Keywords: Hemangioma • External auditory canal

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1. Introduction

Hemangiomas are benign vascular tumors which are common in the head and neck region, but hemangiomas of the external auditory canal (EAC), with or without tympanic membrane involvement, are rare and only individual cases are described in the literature.

So far, a total of 18 hemangiomas of the EAC, with or without tympanic membrane involvement, have been reported in the English language literature. There were 8 cases located in the EAC, so these two case reports are the 19th and 20th cases of hemangiomas of the EAC, and at the same time the 9th and 10th cases located in the EAC alone.

1.1. Case report I

A 38-year-old female patient, with a diagnosis of external auditory canal (EAC) atheroma, made by her otorhinolaryngologist, was admitted to the Emergency Department of the Ear, Nose and Throat Clinic of the Clinical Center of Vojvodina, with a recommendation for atheroma excision. The medical history revealed a month-long feeling of a clogged ear, associated with a sharp pain in the right ear and right-sided occipital headache. The patient received antibiotic therapy, and local therapy with antibiotic ear drops. The therapy showed no significant improvement. Clinical findings were obtained by otomicroscopy and a soft-tissue mass, arising from the EAC, was established. The tumor extended from the right introitus, medially to the lower posterior wall, which almost completely filled the lumen of the EAC; it was smooth on the surface, bluish-pink in color, neither pulsatile nor sensitive to touch. Examination of the EAC was impossible me-

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dially to the tumor and to the tympanic membrane. The otomicroscopic finding of the left ear, anterior rhinoscopy, oropharyngoscopy and indirect laryngoscopy findings were normal. There were no palpable lymph nodes in the neck. There was no tenderness in the region of the mastoid planum, but palpatory tenderness was established in the right intraauricular area. Tuning-fork tests showed the following: Weber test was not lateralized, Rinne test was bilaterally positive, while Schwabach test was bilaterally normal. Otoneurological findings were normal. After the above-mentioned examinations were performed, the patient was hospitalized for further morphofunctional diagnosis with a suspected tumor of the EAC of vascular origin. Due to complete clinical findings, neither puncture nor excision was performed. During the hospital stay, the patient underwent pure tone audiometry (PTA), showing that the hearing threshold was bilaterally normal, whereas tympanometric findings revealed bilateral type-A tympanometric curves. Ipsilateral acoustic stimulation evoked a bilateral stapedius reflex at 0.5 and 1 kHz, at a stimulus level of 100 dB. Morphological diagnosis was made by using computed tomography (CT) scans of the temporal bones, with a series of axial scans before and after intravenous administration of contrast medium. CT-scans revealed an oval, voluminous lesion protruding into the EAC lumen, 17 mm in length, with strong post contrast relatively homogenous high attenuation (Figure 1).

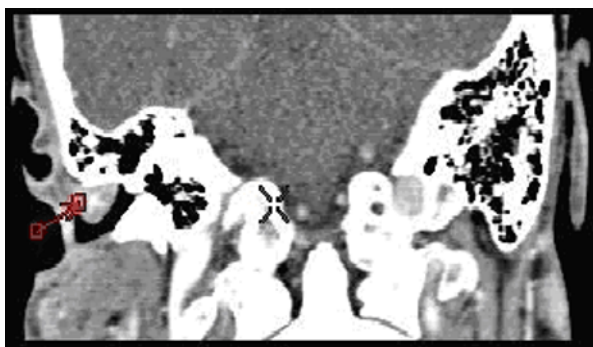


Figure 1. Right temporal bone CT scan with contrast agent showing strong post contrast relatively homogenous high attenuation mass in the External Auditory Canal (EAC).

The lesion was mostly within the bony external auditory canal, with its wider side fixed to the anterior aspect of the mastoid process. The cortex of the overlying region presented with limited bony erosion. The tympanic membrane and cavity, as well as the inner ear presented with normal morphology. The mastoid process was well developed and pneumatized. The morphology of the left temporal bone was normal in all aspects.

The patient underwent the usual preparation for surgery in general anesthesia. Clinical and laboratory findings were normal. Intraoperative otoendoscopy was per-

formed using an endoscope of 0 and 30 degrees. Normal findings of the EAC were found medially to the tumor and to the tympanic membrane. A transmeatal, longitudinal skin incision was made over the lower posterior wall of the EAC, and then a vertical cut along the lateral part of the tumor. Preparation of skin flaps was made, and a complete tumor excision was performed. The excised lesions were sent for histopathological evaluation. Bleeding during the operation was easily managed with cauterization. The posterior bony wall of the EAC towards the mastoid was found superficially eroded, so the surface was smoothed using a diamond burr, until a healthy, smooth surface was obtained. The skin flaps were repositioned and tamponade of the EAC followed. The postoperative course was eventless. Tamponade removal was performed on the 14th postoperative day. The otomicroscopic finding was normal. The histopathological findings revealed cavernous hemangioma (Figure 2).

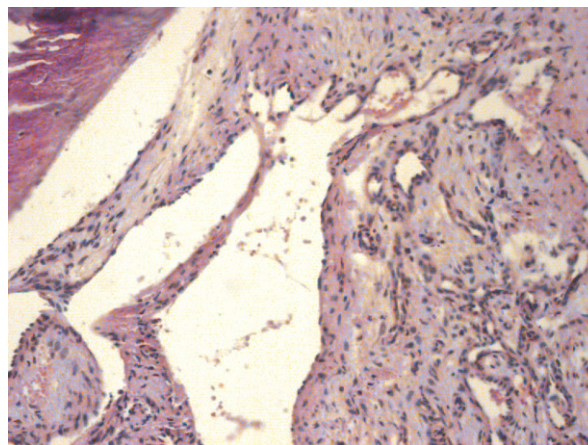


Figure 2. Microscopic section, cavernous hemangioma. Fragment of the bone in the right corner (HE stain, original magnification x200)

The biopsy section of mucosa shows many vascular spaces with blood content. The endothelial cells are flattened with regular nuclei. Most of the vascular spaces are highly dilated. Between the blood vessels, the loose connective tissue is present. The part of hemangioma is close to the bone making the pressure destruction of it.

Five months after surgery, the patient was symptom free. The local, otomicroscopic findings were normal, as well as the control findings of PTA and tympanometry. The patient presented with normal bilateral hearing thresholds, with bilateral type-A tympanometric curves. Magnetic resonance imaging (MRI) of the endocranium also showed normal findings.

1.2 Case report II

A 68-year-old female patient was admitted to the ENT Clinic of the Clinical Center of Vojvodina complaining of

intermittent blood-tinged discharge, right sided otorrhea, impaired hearing and occasional headaches during a period of several months. Glomus tumor was suspected after otoscopy, which revealed a prominent tumor obstructing the right EAC, bluish, smooth-surfaced with mild right preauricular swelling. CT-scans of the temporal bones, without administration of intravenous contrast, showed an isodense area in the right EAC, with signs of chronic changes in the mastoid antrum, without signs of bone erosion. Endocranial MRI findings showed an ovoid, expansive, well-defined soft-tissue lesion, 11x21 x12 mm in size, (AP x LL x CC) (Figure 3); T2W images were hyperintense with intense and relatively homogenous post contrast signal increase, whereas in the subcutaneous periauricular region significant post contrast signal intensity enhancement was recorded. Within the tympanic and mastoid cavities, on the same side, a fluid signal is detected, as well as post contrast mucosal thickening. 3D TOF magnetic resonance angiography (MRA) of the intracranial and neck arteries showed no significant pathologic changes. Audiometric findings showed right-sided, severe, impaired auditory thresholds at frequencies of 0.5-4 kHz, 45-55-70-65 dBnHL, respectively, and a 40-59 dB AB gap, while findings on the left side were normal.

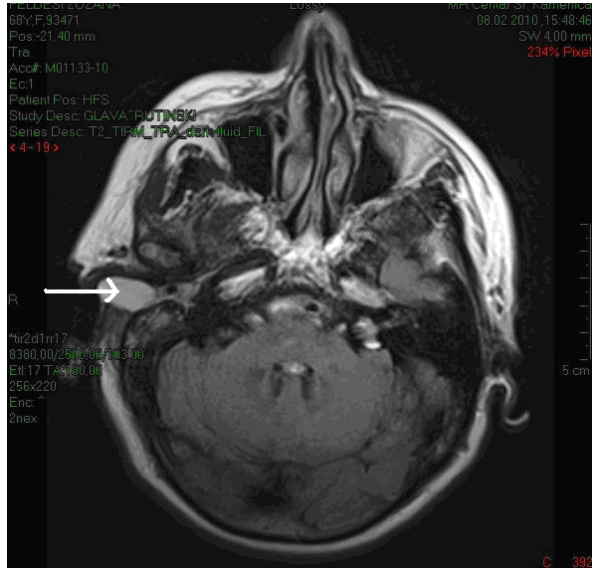


Figure 3. MRI showing an ovoid, expansive, well-defined soft-tissue lesion, 11x21 x12 mm in size, (AP x LL x CC) in the right EAC.

After all clinical and morphofunctional findings were obtained and the opinion of an internist was sought, the patient underwent surgery under general endotracheal anesthesia. A retroauricular approach was used for complete tumor excision. The tympanic membrane was not involved. However, a central tympanic membrane perforation was detected, as well as hyperplastic muco-

sa in the tympanic cavity with intact ossicular chain. The patient underwent myringoplasty. The postoperative course was eventless. The histopathological findings revealed a mixed cavernous and capillary hemangioma in the biopsy of mucosa with non-keratinized stratified squamous epithelium at the surface. A great number of blood vessels are in the whole lamina propria. Vascular spaces are with thin walls, with various luminal diameter and flattened endothelial cells. In some vessels blood cells are visible. In an area (not shown at the picture) recent thrombi are in the blood vessels with the ulceration of the surface mucosa (Figure 4).

Three months after surgery, the otomicroscopic finding was normal, while the PTA findings were unchanged.

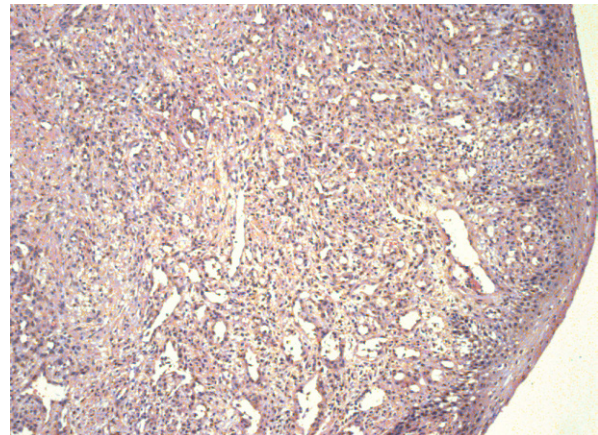


Figure 4. Histopathological findings revealed a mixed cavernous and capillary hemangioma (HE stain, original magnification x100)

2. Discussion

Hemangiomas of the external auditory canal (EAC) are rather rare. To our knowledge, literature review revealed that only 18 cases of hemangiomas of the EAC, with or without involvement of the tympanic membrane or the tympanic cavity, have been reported so far (Medline search, 1980 to present). Out of these 18 reported cases, in 8 cases hemangiomas were located only within the EAC (Table 1) [1-8].

The first case of a cavernous hemangioma located only in the EAC was reported by Hawke and von Noststrand in 1987, and after that, in 2009, the last reported cases of hemangiomas at the same location were reported by Rutherford. Thus, these two case reports are the 9th and 10th cases of EAC hemangiomas reported so far. The age of patients ranges from 31 to 72 years, with mean age of 55.7 years. The male to female ratio is 1:2.3, respectively, whereas the male to female ratio of all reported cases of EAC hemangiomas, with and

Table 1. Hemangiomas of the external auditory canal – literature review.

	Author	Year	Age/Gender	Location/side	Pathology	Management
1.	Hawke <i>et al.</i>	1987	55/M	EAC/R	Cavernous	Transcanal excision
2.	Limb <i>et al.</i>	2002	67/F	EAC/L	Cavernous	Endaural excision
3.	Reeck <i>et al.</i>	2002	53/M	EAC/L	Cavernous	Endaural excision
4.	Rehan <i>et al.</i>	2003	67/F	EAC/L	Cavernous	Endaural excision
5.	Yang <i>et al.</i>	2006	72/F	EAC/L	Cavernous	Transcanal excision
6.	Verret <i>et al.</i>	2007	31/M	EAC/R	Cavernous	Postauricular excision
7.	Covelli <i>et al.</i>	2007	45/F	EAC/L	Cavernous	Endaural excision
8.	Rutherford <i>et al.</i>	2009	62/F	EAC/L	Cavernous	1. Postauricular excision 2. Transcanal excision, TMJ arthrotomy
9.	Vlaški <i>et al.</i>	2010	38/F	EAC/R	Cavernous	Transcanal excision
10.			68/F	EAC/R	Capillary/ Cavernous	Postauricular excision

m=male; *f*=female; EAC=external auditory canal; TMJ=temporomandibular joint, R=right; L=left

Table 2. Findings of pure tone audiometry (PTA) and computed tomography (CT) of the temporal bones in patients with EAC (external auditory canal) hemangiomas.

	Author	PTA	Temporal bone CT
1.	Hawke <i>et al.</i> 1987	Mixed hearing loss, AB gap 10-30 dB	A soft tissue mass in posterior-inferior aspect of the EAC
2.	Limb <i>et al.</i> 2002	Moderate mixed hearing loss	15 mm nonenhancing mass lateral to the tympanic membrane
3.	Reeck <i>et al.</i> 2002	Mixed hearing loss, AB gap 10-30 dB	An isolated soft tissue mass in posterior-inferior aspect of the EAC
4.	Rehan <i>et al.</i> 2003	Moderate to profound mixed hearing loss	Radiography- intact tympanic membrane middle ear normal
5.	Yang <i>et al.</i> 2006	Mild to moderate SN hearing loss	A small soft tissue mass in the EAC
6.	Verret <i>et al.</i> 2007	Mild conductive hearing loss	16 mm mass in the EAC, limited bony erosion of the EAC
7.	Covelli <i>et al.</i> 2007	Mild conductive hearing loss	15 mm soft tissue mass in the EAC lateral to the tympanic membrane
8.	Rutherford <i>et al.</i> 2009	Normal pure tone thresholds	Normal temporal bones
9.	Vlaški <i>et al.</i> 2010	Normal PT threshold	17 mm soft tissue mass in posterior-inferior aspect of the EAC, limited bony erosion
10.		Moderate to profound mixed hearing loss	An isodense area in the EAC, with sings of chronic changes in the mastoid

AB= air bone; SN= sensorineural

without tympanic membrane or cavity involvement, is 1:1. The symptoms of EAC hemangiomas vary: from asymptomatic cases, to cases with otorrhagia, ear canal obstruction, hearing impairment and earache. Six out of eight thus far reported cases of EAC hemangiomas, affected the left ear, while in our case reports hemangiomas were located in the right ears.

Clinical and otomicroscopic examinations of EAC hemangiomas show that they are purple, soft-tissue and exophytic masses. In regard to their size, it varies from 2 mm to 29 mm. In these two case reports the lesions were 17 mm and 21 mm in size.

Apart from history taking, the diagnosis is made based on clinical otorhinolaryngologic and otomicroscopic examinations, including functional diagnosis using PTA and morphological examination using CT of the temporal bones (Table 2).

Endocranial MRI was used only in one reported case of EAC hemangioma, by Rutheford in 2009. Due to postoper-

ative otorrhagia, which occurred on the 25th postoperative day, reoperation was done using transmeatal approach with arthrotomy of the temporomandibular joint [1]. Computed tomography of the temporal bones is considered to be the most accurate diagnostic procedure for hemangiomas of the EAC. In the above-mentioned case report of Rutherford (2009), the initial CT of temporal bones did not show the complete extension of the hemangioma, thus MRI with gadolinium contrast showed the spread of the lesion into the mandibular fossa. In our second case report, MRI was performed prior to surgery, due to clinical findings of otoscopy: prominent bluish lesion with preauricular swelling and occasional episodes of blood-tinged otorrhagia; MRA also showed no significant pathological changes, both of great importance in planning surgical therapy. Angiography, generally speaking, is not necessary, except in cases where extensive hemorrhage may be expected [1].

The final diagnosis is based on histopathological (HP) findings. Histopathological types of EAC heman-

giomas include: capillary, cavernous, and mixed type hemangiomas. Out of the 18 so far reported cases of EAC hemangiomas, 17 had reported the following HP findings: cavernous type was found in 76.4% of patients, capillary in 17.6%, and mixed type in 5.8% of cases. Our first case report is about a patient who presented with the most frequent type – cavernous hemangioma, whereas the second had a mixed type hemangioma.

Therapeutic modalities vary, from follow-up of patients, to partial temporal bone resection [9]. In cases where follow-up is performed, patients mostly present with small, capillary EAC hemangiomas. In 1978, after 18 months of follow-up, Balkany found a significant, two-fold increase in the size of the tumor, and it was an indication for surgery. Local, therapeutic excision was performed using one out of the following three approaches: transmeatal, endaural or retroauricular in 13 out of 18 cases; tympanoplasty was performed in 3, while two patients presented with recurrences. In 1990, Jackson performed a partial temporal bone resection, whereas in 2009, Rutherford performed a reoperation using transmeatal excision with arthrotomy of the temporomandibular joints, but with previously performed blood vessel embolization [1]. In our first case report, local excision of the hemangioma was performed using a transmeatal approach, and 5 months after surgery, the patient presented with normal local, otomicroscopic and MRI findings of the endocranium. In the second case, postauricular approach was used for the excision of the

hemangioma, but intraoperatively, a central type perforation of the tympanic membrane was detected, and myringoplasty was performed. Three months after the surgery, the patient presented with normal otomicroscopic and PTA findings unchanged.

3. Conclusion

Hemangiomas of the external auditory canal are rare conditions. In the morphological diagnosis, computed tomography of the temporal bones is the leading diagnostic modality, whereas intraoperative otoendoscopy helps to evaluate the medial part of the external auditory canal and the integrity of the tympanic membrane in relation to the tumor, which limits otomicroscopy due to its size. Biopsy of vascular tumors is not recommended, because it may cause excessive bleeding. Complete surgical excision of cavernous hemangiomas of the external auditory canal is the therapy of choice, with minor risk of hearing damage. Endocranial magnetic resonance imaging is valuable in the evaluation of the spread of the entire lesion, as well as in the postoperative follow-up.

Conflict of interest

The authors received no fundings from any organization, none of the authors has any conflict of interest, financial or otherwise.

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