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Case Report

Cavernous Hemangioma of the Mastoid Antrum

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Hemangioma is a common vascular neoplasm that arises in the head and neck regions but is rare in the petrous bone. We report the first case of a solitary cavernous hemangioma in the mastoid antrum. A 68-year-old woman visited our hospital with a complaint of tinnitus without any other symptoms. Tinnitus of the right ear occurred especially when the patient yawned or swallowed. Both tympanic membranes appeared normal on otoscopic examination. On pure-tone audiometry, mild hearing loss up to 25 dB was detected in the right ear. Temporal bone computed tomography revealed a 7.0 mm × 4.5 mm × 5 mm, solitary soft tissue mass in the aditus ad antrum. Excisional biopsy was performed under general anesthesia through the canal wall as in a mastoidectomy. The mass was completely removed without any bleeding or ossicular chain damage. The mass was confirmed as a cavernous hemangioma. During follow-up, the patient's tinnitus and right low-tone hearing loss improved. No solitary hemangioma of the mastoid antrum has been reported previously. Surgical excision of the lesion appears to be proper treatment to achieve pathologic confirmation along with resolution of symptoms.

KEYWORDS: Cavernous hemangioma, mastoid antrum, middle ear surgery

INTRODUCTION

Hemangioma is a very common benign tumor that may occur in the head and neck region and mostly appears as a cutaneous lesion. More than 50% of hemangiomas arise in the head and neck area; 95% of hemangiomas are present by the age of 6 months, whereas the prevalence decreases to 1.6% by age 5 years, with most lesions resolving without treatment. Spontaneous degeneration can occur until the age of 12, and hemangiomas that have not degenerated by this time likely will not resolve on their own.[1] Solitary lesions comprise 80% of diagnosed tumors, and more females than males are affected at a ratio of 3 : 1.[2] Hemangiomas are classified as capillary hemangioma or cavernous hemangioma according to pathological findings.

These tumors are not frequently reported in the petrous bone, including the middle ear. Until recently, only 31 cases of hemangioma have been documented in the external auditory canal (EAC) and middle ear cavity (MEC). No cases have been reported in the mastoid cavity alone, and 26 cases were pathologically confirmed. Among the existing cases, 65% (17/26) were cavernous hemangiomas. In addition, 38% of reported cases occurred in the EAC (12/31), while 26% of reported cases were on the tympanic membrane (TM; 8/31). Cases in both the EAC and TM accounted for 19% of the total (6/31). Four cases occurred in the EAC/TM/MEC, and there was also 1 case in the TM/EAC that included bone.[3] To our knowledge, hemangioma in the mastoid antrum alone has not been previously reported. We report a case of cavernous hemangioma that occurred only in the mastoid antrum and was surgically removed.

CASE PRESENTATION

A 68-year-old woman visited our outpatient clinic complaining of tinnitus of the right ear for the past few months. The tinnitus sounded "clattering" when the patient yawned or swallowed. She had no symptoms of ear discharge or otalgia and no history of facial palsy or vertigo. There were no abnormal otoscopic findings in either the TM or EAC. Pure-tone audiometry detected a threshold of 20 dB at 500 Hz, 25 dB at 1 kHz, 25 dB at 2 kHz, and 25 dB at 4 kHz, without an air-bone gap, consistent with presbycusis. Impedance audiometry showed type A. Additional high-resolution non-enhanced computed tomography (CT) scanning of the temporal bone was performed to evaluate anatomic abnormalities of the middle ear and revealed a 7.0 mm × 4.5 mm × 5 mm, well-delineated, rounded tissue shadow at the right aditus ad antrum (Figure 1). Computed tomography findings showed that the mass was in contact with the short process of the incus. However, there was no dislocation or erosion of the ossicular chain. Surgery was planned

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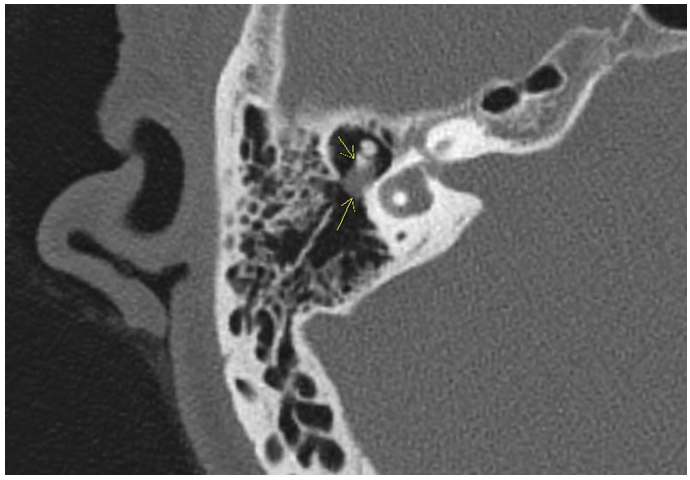

Figure 1. Preoperative computed tomography showed a round, high soft tissue density mass in the mastoid antrum that was in contact with the short process of the incus.

for pathological diagnosis, and excisional biopsy was performed via the canal wall up mastoidectomy. The antrum was exposed through the canal wall up mastoidectomy, and the mass was located in the aditus ad antrum. Under surgical microscopy, the mass was bright reddish and round and was in weak contact with the short process of the incus (Figure 2). No noticeable feeding vessels for the mass were identified. Complete mass removal was possible using microinstruments without problematic bleeding or any ossicular damage. The mastoidectomy wound was repaired in the usual manner.

Histopathologically, the lesion was diagnosed as a cavernous hemangioma (Figure 3). The patient recovered uneventfully. During follow-up, tinnitus completely resolved. The postoperative pure-tone audiometry slightly improved to 15 dB at 500 Hz compared to 20 dB before surgery. This study was approved by Ethics Committee of

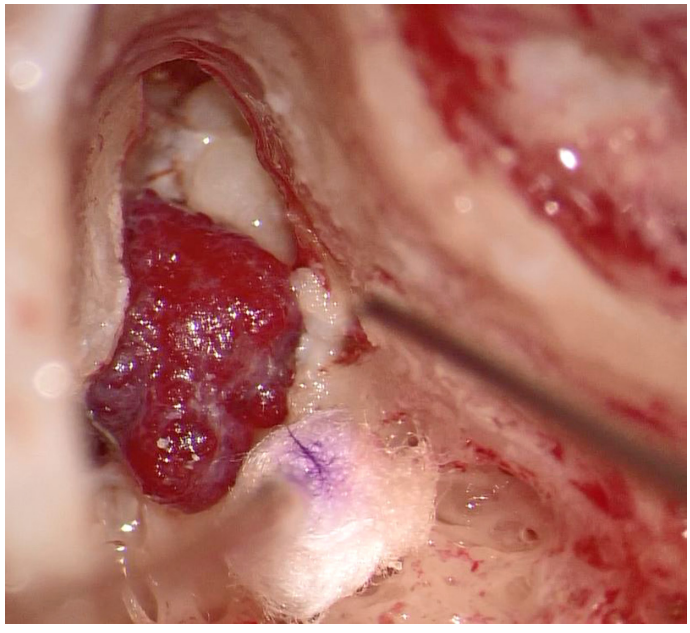

Figure 2. Clinical appearance of the hemangioma following canal-wall-up mastoidectomy. A bright reddish, round mass was identified in the aditus ad antrum and was in weak contact with the short process of the incus.

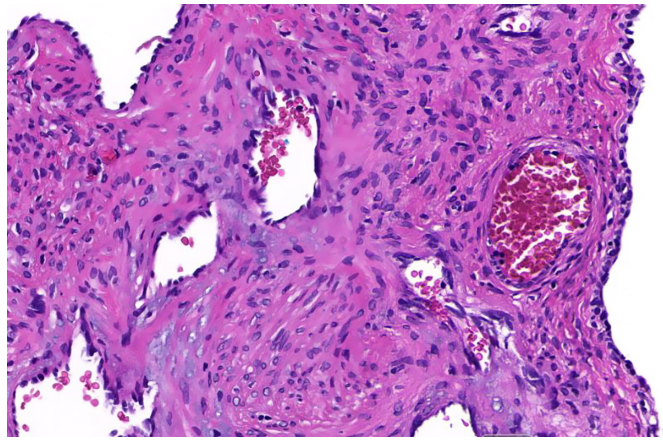

Figure 3. Pathologic findings revealed large dilated vascular spaces filled with red blood cells, consistent with cavernous hemangioma.

Catholic University (Approval No: PC23ZASI0013; Date: February 12, 2023). Written informed consent was obtained from the patients who participated in this case.

DISCUSSION

Hemangioma is a common vascular neoplasm that occurs in the head and neck regions. However, hemangioma in the temporal bone is rare. Symptoms of otologic hemangioma vary depending on location of the tumor from local inflammatory reactions such as otalgia or otorrhea; to mass effects such as pain, aural fullness, and tinnitus; to sensorineural hearing loss or facial palsy. Considering that hemangiomas are a type of vascular tumor, ear bleeding may occur. In our case, clicking tinnitus upon swallowing or yawning was the patient's only symptom and improved after surgical mass removal. This change indicates that the clicking tinnitus was caused by physical friction of the mass adjacent to the short process of the incus.

As in most other cases, radiological evaluation (such as high-resolution CT and gadolinium-enhanced magnetic resonance imaging (MRI)) is essential to determine the diagnosis and treatment strategy. Differential diagnosis might include other vascular lesions such as a glomus tumor or an aberrant carotid artery, a high jugular bulb, carcinoma, melanoma, etc.[4,5] The location, size, bone erosion, and patient symptoms should also be considered when establishing a treatment plan. Preoperative embolization is not generally necessary,[6] but a few cases require intervention according to size, location, and growth tendency. Cases of operation after embolization for suspected feeding vessel involvement have shown good results. Yosaku Torii et al[7] reported a case of a hypervascular lesion that was successfully resected after preoperative embolization.[7]

Hemangioma is histologically divided into capillary and cavernous types and can occur wherever blood vessels are present.[8] Capillary hemangiomas are found in the skin and subcutaneous lesions, while cavernous hemangiomas are often identified in elderly patients in deeper tissues such as the larynx, muscles, liver, and brain. Cavernous hemangioma has not been frequently reported in the petrous bone, including the middle ear. Pathologic specimens of both types have shown numerous vascular structures containing red blood cells or transudate with a preserved normal, single endothelial cell layer. The difference between capillary hemangioma and cavernous hemangioma

is the presence of vessel dilatation. Several immunohistochemical stains are used for diagnostic markers, such as CD31, CD34, factor VIII-associated protein, vascular endothelial growth factor (VEGF), and others, among which GLUT 1 is the most useful and widely used marker.[2,9]

Hemangioma has a unique developmental course in that the early proliferation phase is followed by spontaneous involution. The pathogenesis of hemangioma has not been clearly identified; the aforementioned features were probably due to overexpression of growth factors (such as fibroblast growth factor and VEGF) during proliferation and an increased level of tissue metalloproteinase during the involuting phase.[2,8]

Considering the developmental progression, the treatment plan should be tailored to each patient. Close follow-up could be an appropriate management method in asymptomatic patients.[2,10] Laser therapy can be used to treat hemangiomas located in superficial sites.[10] Surgical treatment can be performed if the patient is compliant. If not, although there is no established dose for adults, medical treatment such as systemic steroid or beta-blocker might be an alternative.[2,9,10] In our case, MRI/embolization was not performed preoperatively because the mass was small and solitary and was confined to the antrum.

CONCLUSION

Hemangioma only in the mastoid antrum alone has not been previously reported. Contrary to other cutaneous hemangiomas, surgical excision is frequently the treatment of choice for temporal bone hemangioma.

Ethics Committee Approval: This study was approved by Ethics Committee of Catholic University (Approval No: PC23ZASI0013; Date: February 12, 2023).

Informed Consent: Informed consent was obtained from the patient who agreed to take part in the study.

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