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Acquired Capillary Hemangioma With Features of Tufted Angioma in the External Auditory Canal

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Abstract: Tufted angiomas may occur in the head and neck region, but the external auditory canal is a previously undefined localization. There are only 19 cases of hemangiomas reported in this unique localization. However, this case is the first capillary hemangioma of the tufted variant being reported with a recurrence after 7 years after surgical excision in a 47-year-old male patient.

Key Words: Hemangioma, tufted angioma, external auditory canal

Hemangiomas are the most common benign vascular neoplasm in the neonatal period. Incidence of this disease is 1% to 2.6% at birth and 10% in the first 1 year of life. Fifty percent of these lesions regress generally within during the first 5 years of life and the rest usually by the age of 10 to 12 years. The management of this disease is mainly conservative because these lesions usually involute spontaneously.¹

There are 2 major types of hemangiomas: cavernous and capillary. The capillary type is lobulated and has no capsule. The color is purplish and infiltrates the surrounding tissues. Solid masses of cells and multiple anastomizing capillaries are seen microscopically. The cavernous type is formed by dilated blood vessels or sinusoids lined by endothelium.² The characteristic histologic feature of the acquired tufted angioma, which is a special form of capillary hemangioma, is the formation of clustered capillaries.³ Tufted angioma was first recognized as an entity in the English-language literature in 1976,

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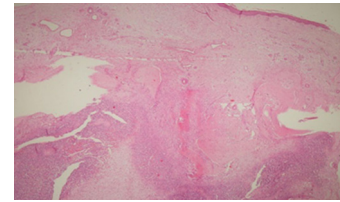


FIGURE 1. Computed tomographic imaging of the mass localized in the external auditory canal.

although identical cases had been previously described in the Japanese literature as “angioblastom.”⁴

CLINICAL REPORT

A 47-year-old man came to the ear, nose, and throat department with the complaint of hearing loss in the right ear. He had a history of an operation for mass in this area 7 years ago, and acquired tufted angioma was detected pathologically. In the examination of the affected ear, we found a nodular mass lesion in the posterior superior region of the right external auditory canal. Conductive air hearing loss (20–25 dB air bone gap at 250–4000 Hz) were detected in the same ear otologically. Magnetic resonance imaging (MRI) and computed tomographic (CT) studies revealed a 2-cm mass in the external ear canal, with contrast enhancement (Figs. 1 and 2). Transcanal mass excision surgery was performed, and microscopic examination showed a vascular lesion with infiltrating features of capillary hemangioma like nodules resembling “cannonballs,” which are descriptive features of tufted angioma (Figs. 3 and 4). The tissue removed was negative for neuroendocrine cancer markers, that is, synaptophysin, chromogranin, and neuron-specific enolase. Reticulin stain revealed a vascular pattern, and CD34 antibody, some vascular spaces scattered throughout the nodular masses. The previous excision 7 years before showed the same morphology. The patient healed immediately after surgery, and no evidence of tumor recurrence was detected in the postoperative period of 1 year.

DISCUSSION

Hemangiomas are benign vascular tumors that can be seen in the head and neck region⁵ and are encountered typically by age of 1 year and involute by age of 5 or 6 years in children.⁶

The external auditory canal is a rare site with 19 cases being previously reported in the literature. Capillary hemangiomas were detected in only 4 of these 19 cases histopathologically. Our case is the first defined case of hemangioma with an acquired tufted angioma pattern in the external auditory canal.

Hemangiomas of the external auditory canal and/or tympanic membrane are extremely rare and appeared dominantly between the sixth and seventh decades of life.⁷ This disease may lead to otorrhea, blood-tinged otorrhea, aural fullness, and tinnitus and hearing loss

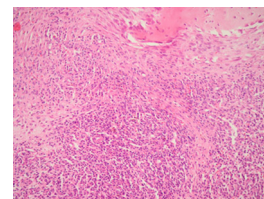


FIGURE 2. Magnetic resonance image with gadolinium revealing the mass lesion.

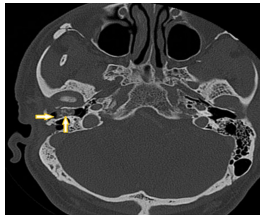


FIGURE 3. Low-power view of the excised mass showing cellular nodules resembling “cannonballs” within osseous fragments (hematoxylin and eosin; original magnification, $\times 100$).

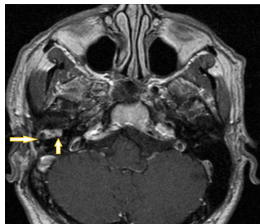


FIGURE 4. Higher-power view showing oval or spindle cells with slit-like luminal spaces (hematoxylin and eosin; original magnification, $\times 200$).

symptoms but may be found incidentally in the asymptomatic cases. With the otoscopic examination, a purple, red, or violaceous vascular mass may be seen in the external auditory canal.^{6,8}

Although hemangiomas are benign tumors, recurrence may be seen because these lesions are not capsulated and have infiltrative features. The recurrence in our patient may be due to the infiltrative neoplastic cells remaining among bone lamellas.

Differential diagnoses include attic cholesteatoma with aural polyp, glomus jugulare, granulation tissue, arteriovenous malformation, and carcinoma of the external auditory canal. Pure tone audiometry, CT, and/or MRI may be used in diagnostic evaluation.⁹ Computed tomographic scan is the first choice for detecting the size of the lesion and amount of bone infiltration. The exact localization of the tumor and degree of soft tissue involvement is very difficult with MRI because air and bone structures appear black.

The treatment of this lesion is complete excision. The small lesions of external auditory canal, similar to our case, might be treated with transcanal excision.⁸ Large lesions extending to the tympanic membrane and mastoid bone might require tympanoplasty, mastoidectomy, or partial temporal bone resection.^{6,10} The most important pathologic differential diagnosis in a male adult patient is nodular Kaposi sarcoma, from which tufted angioma is distinguished by its “cannonball” pattern, lack of a significant spindle cell population, and vasoformative reticulin pattern.⁴

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Assessment of the Epidemiological Profile of Patients With Dentofacial Deformities Who Underwent Orthognathic Surgery

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Abstract: The present study aimed to establish the profile of patients who underwent orthognathic surgery in a private clinic by evaluating their demographic characteristics, their facial types, and aspects related to the surgical procedures that were performed. The sample consisted of 419 medical records from male and female patients aged 15 to 62 years who underwent orthognathic surgery between 2001 and 2011. A single examiner collected data by evaluating a database of information extracted from medical records, particularly radiographic and photographic analyses. The following criteria were evaluated: gender, age, skin color, type of orthognathic surgery, type of associated temporomandibular joint (TMJ) surgery, complications, and recurrences. Seventeen patients were rejected because they had incomplete records. The average age of the patients was 28.5 years old; most were females (255 patients) and faioderm (295 patients). The most prevalent facial pattern was Pattern III (n = 166, 41.3%). Orthognathic surgery that affected the maxilla, jaw, and chin was the most prevalent type (n = 199, 49.5% of cases).

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