Capillary haemangioma of external and middle ear

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ABSTRACT

Haemangiomas are relatively common in the head and neck region, but cases in the temporal bone are rare. Temporal bone haemangiomas tend to arise from the geniculate ganglion, internal auditory canal and the beginning of chorda tympani due their vascularity. Haemangiomas originating from the external canal and middle ear are exceedingly rare. Due to the very limited data, the true natural history of this peculiar entity remains unknown. Here we report a case of capillary haemangioma involving the external and middle ear and discuss the diagnosis, investigative work up and treatment of the disease.

Keywords: Capillary haemangioma, middle ear, external ear, vascular tumour

INTRODUCTION

Haemangiomas are relatively common in the head and neck region, but cases in the temporal bone are rare. Temporal bone haemangiomas tend to arise from, by decreasing order in frequency, the geniculate ganglion, internal auditory canal and beginning of chorda tympani due their vascularity. Haemangiomas originating from external canal and middle ear are exceedingly rare. The true natural history of this peculiar entity remains unknown. We report a case of capillary haemangioma involving the external and middle ear.

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CASE REPORT

A 50-year-old man with background history of hypertension presented with persistent left ear serous discharge, and episodic ear bleeding for a month. This was associated with pulsatile tinnitus. No facial nerve dysfunction. Otoscopic examination revealed a mass occupying the entire left bony ear canal resembling an ear polyp. Pure tone audiometry revealed moderate to profound mixed hearing loss on the left side, and mild to severe sensorineural hearing loss on the opposite side. His condition failed to improve despite a course of antibiotic and this prompted the referring doctor to biopsy the mass. Histology revealed a capillary haemangioma.

A high-resolution computed tomogra-

tomography (CT) scan of the temporal bone showed a soft tissue mass in the left external auditory canal and middle ear with evidence of left mastoiditis. Erosion was also seen affecting the floor of left ear canal. A contrast enhanced magnetic resonance imaging (MRI) (Figure 1) revealed the enhancing soft tissue mass to affect part of the mastoid air cells and the left petrous apex. The inner ear structures were normal and there were no intracranial extension noted. Cerebral angiogram revealed enhancement on the left temporal region during early arterial phase. Seventy per cent of the vascular supply supplied by the branches of the left external carotid artery was successfully embolised.

At surgery, the external auditory canal was truncated via post auricular approach and periosteal flap. A canal wall down mastoidectomy was performed, revealing the vascular tumour in the mastoid antrum and attic extending down to the mesotympanum and hypotympanum. The Eustachian tube opening was obliterated. Dehiscence of the tympanic facial nerve was noted. The ossicles were intact. The haemangioma was radically resected with bipolar diathermy and cold instruments. Blood loss was minimal. Histopathological examination confirmed the clinical diagnosis of capillary haemangioma. He was well and there was no evidence of recurrence on follow up.

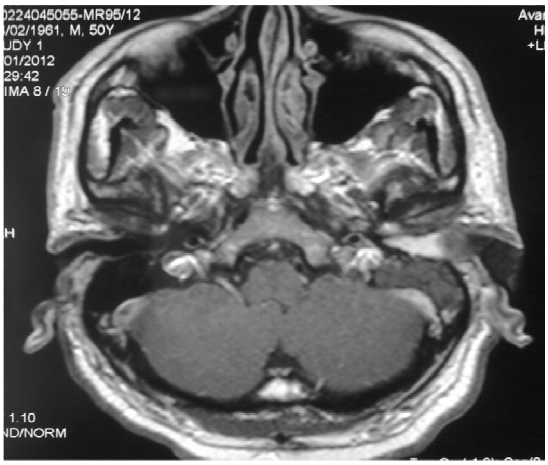

Fig. 1: Axial view of contrasted MRI revealing nature and extent of the lesion in the left external and middle ear.

DISCUSSION

Capillary haemangioma in the middle ear was first described by Jones in 1930. [1] Mangham *et al.* reported that haemangiomas accounted for 0.21% a large series (n=1,430) intratemporal tumours. [2] Up to now, only 18 cases have been reported in the English literature. [3, 4, 6-10]

Temporal bone haemangiomas tend to arise from the geniculate ganglion, the internal auditory canal and beginning of chorda tympani due their vascularity. [4, 5] The most common presenting features, in the order of decreasing frequency, are conductive hearing loss, pulsatile tinnitus, bloody otorrhoea, otalgia, and otitis media. [5] It can be easily mistaken for other pathologies, due to the non specific symptoms and also its rarity. Differential diagnosis to be considered includes other vascular lesions (glomus tumour being the most common vascular tumour in the middle ear, high jugular bulb, aberrant intratympanic tympanic vessel, arteriovenous malformation), neoplastic lesions (meningioma, rhabdomyosarcoma, carcinoma, melanoma, pyogenic granuloma) or inflammatory lesions which are far more prevalent (cholesterol granuloma, ear polyp). [11] In our, the patient was initially treated for chronic otitis media

based on the history and otoscopic findings. It was only after a biopsy that the diagnosis was revealed. Retrospectively, a high index of suspicion is absolutely needed to diagnose this condition clinically.

Literature review revealed that almost all reported cases suggest that high-resolution CT scan and MRI as useful in evaluating this vascular tumour. CT scan provide a good assessment of the extent and bone MRI angiography the nature of the mass more definitely. However, one should bear in mind that MRI features of glomus tumours may resemble those of haemangioma. [12] Cerebral angiography helps to demonstrate the origin of the vascular tumour and embolisation could be done at the same sitting. The role of pre-operative embolisation in reducing intra-operative blood loss has been well established. However, the afferent vessels may not be identified. [4, 10] In our patient, bleeding has probably reduced by the embolisation.

The treatment for this condition remains surgery. All the 18 reported cases were treated with surgery except for two cases. One had a spontaneous involution [5] and the other was treated with radiotherapy. [6] The natural history of haemangioma in other head and neck region involves spontaneous involution. Hecht *et al.* postulated that age of onset may be predict the possibility of spontaneous regression. [5] However, haemangioma in the middle ear is slightly different from other regions. It occupies a small, enclosed cavity and cause symptoms at early stage and one may not want to wait until complications arise. Generally, most advocate early resection as advocated. In our patient, after embolisation, a canal wall down procedure was performed to achieve a superior access to the tumour and better control of the bleeding.

Apart from surgery, there are interesting development in alternative treatment modality. Kostrzewa *et al.* described the use of CO_2 laser-assisted excision of tumour which reduced the risk of bleeding and allowed better visualisation. [7] Pavamani *et al.* reported the first time use of radiotherapy as the primary treatment of an inoperable capillary haemangioma of external and middle ear with a five years recurrence-free period. [6] recent, propranol has been used extensively in treating haemangiomas in other body regions. However, its use in intratemporal haemangioma is limited by the low prevalence of such disease. All these alternatives are still relatively new and lack of good evidence to support their usage. The risk of irradiation-induced malignancy has to be considered if primary radiotherapy were to be offered. These modalities might have a role to treat this condition conservatively, but further research and study would be required to establish their efficacy. Since surgery has given a good success rate with minimal morbidity, alternative modalities should only be applied to selected cases.

In conclusion, capillary haemangioma of external and middle ear is rare, and a high index of suspicion is required for early detection. The aetiology and pathogenesis are still not fully understood. Every reported case will increase the awareness among the clinicians. This will contribute to the formulation of a universally accepted guideline on the management protocol in future. Until then, each and every case has to be reviewed and managed on individual basis.

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