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Lobular Capillary Haemangioma Of The Middle Ear- A Rare Case Report.

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Abstract
'Lobular Capillary Haemangioma' of middle ear is a rare, benign disease. It is difficult to diagnose clinically. Here we report a case of a 37-year-old female who presented with the symptom of gradually progressive hearing loss, earache and perception of buzzing sound in the right ear for one and a half years. On examination, a red mass of the size of a pea-nut was found in the middle ear on the right side. The mass was pushing the tympanic membrane outwards. The mass was excised using anterior tympanotomy approach and subsequently diagnosed as 'Lobular Capillary Haemangioma' on histopathological examination.
Keywords: *Lobular Capillary Haemangioma, Middle Ear, Tinnitus.*

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I. Introduction:

Haemangiomas are benign vascular lesions and can develop in any region of the body including the head and neck. 'Lobular Capillary Haemangioma' is a benign vascular tumour derived from the capillaries. It is different from capillary haemangioma by certain features like it is lobulated, sessile, smaller in size, less sexual preponderance etc. It may develop at a number of sites in the body including gastro-intestinal tract,[1] sinonasal area,[2] thigh region [3] etc. Topdag et.al. [4] reported a case of 'Lobular Capillary Haemangioma' in the external auditory canal. Barati et. al. [5] and Varshney et.al. [6] reported cases of capillary haemangiomas in the external auditory canal. Pistorio et. al. [7] and Nouri et. al. [8] reported cases of capillary haemangiomas in the middle ear. There are a few case reports of capillary haemangiomas involving the external auditory canal and the middle ear, extending towards the mastoid area. [9, 10, 11] But we have not found any case report of 'Lobular Capillary Haemangioma' in the middle ear. When it is present in the external auditory canal, it initially presents as a small vascular lesion in the deep posterior bony external auditory canal or over the posterosuperior tympanic membrane. It is usually asymptomatic until it becomes bigger or bleeds due to the manipulation by the patient. As 'Lobular Capillary Haemangioma' is rare in the middle ear, there is lack of knowledge regarding its clinical presentation. Diagnosis and management of such cases often poses problems.

The differential diagnosis includes paraganglioma, high jugular bulb and adenomatous tumour. Some haemangiomas may regress spontaneously, while others continue to grow and are locally destructive. Surgical excision is the treatment of choice in these cases. But as some tumours may regress spontaneously, an expectant approach may be appropriate, particularly if there is apprehension of compromise of middle ear function after surgery. The excised specimen should be sent for histopathological examination to confirm the diagnosis.

II. Case Report:

A 37-year old female from a low socio-economic stratum residing in a rural area of West Bengal, India, presented with the complaints of gradually progressive hearing loss, earache and perception of buzzing sound in the right ear. She had been having these symptoms for a year and a half. She had visited multiple doctors but her condition did not resolve. The pain was intermittent and throbbing in nature, radiating to right neck and shoulder. There was no history of any discharge or bleeding from the same ear.

On examination, a reddish mass was seen deep to the tympanic membrane on the right side. On otoscopic examination, the same mass was found to be pushing the tympanic membrane outwards. The tympanic membrane was congested but intact. Cone of light was absent but no pulsation was noted over the mass. The patient was then taken to the minor ENT-OT and oto-endoscopic examination was done (Fig.-1) to confirm the findings. The cone of light was shifted to the postero-superior quadrant and a small retraction pocket was visible in the antero-superior quadrant. Examination of the other parts of the body revealed no other abnormality.

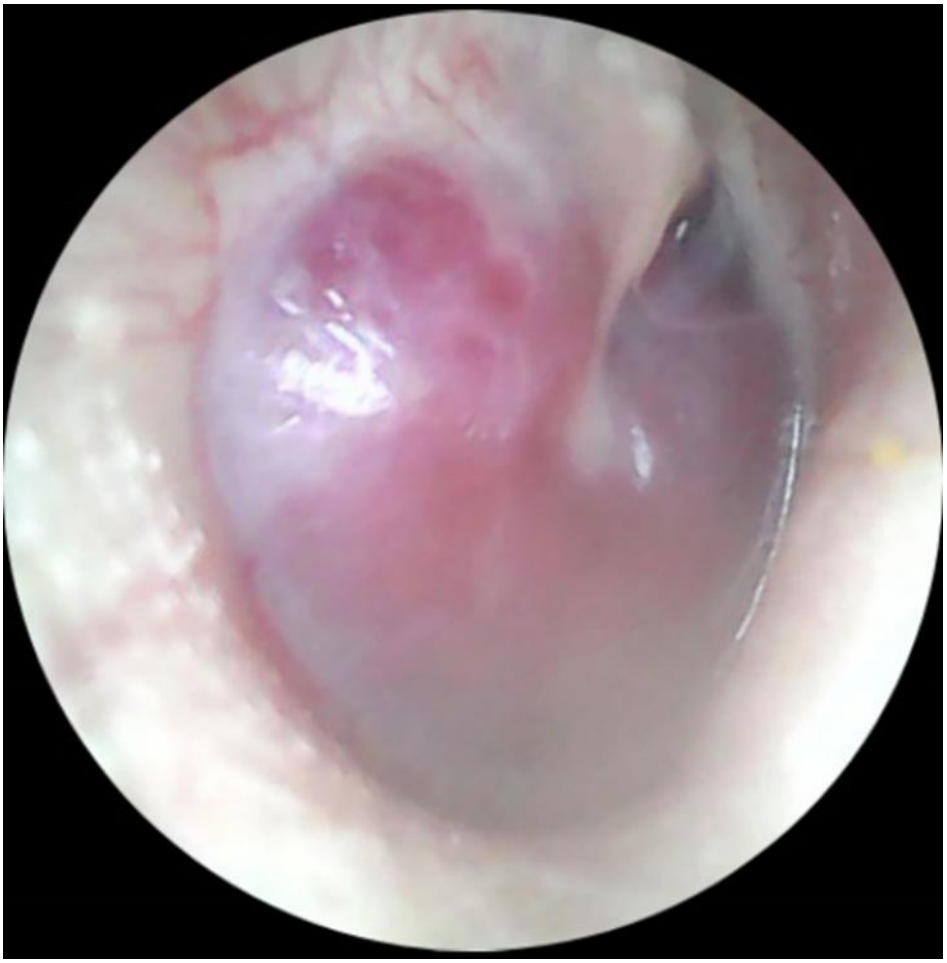

Fig. 1:- Endoscopic image of the reddish, bulging mass deep to the tympanic membrane.

On pure tone audiometry, an average hearing loss of 28.3 dB in the right ear and 21.6 dB in the left ear was found.

In non-contrast HRCT of temporal bones (Fig- 2 & 3), a focal soft tissue dense area measuring 7mm x 7mm was found in between the right tympanic membrane and the promontory, extending to the adjacent areas. The ossicles were found to be intact.

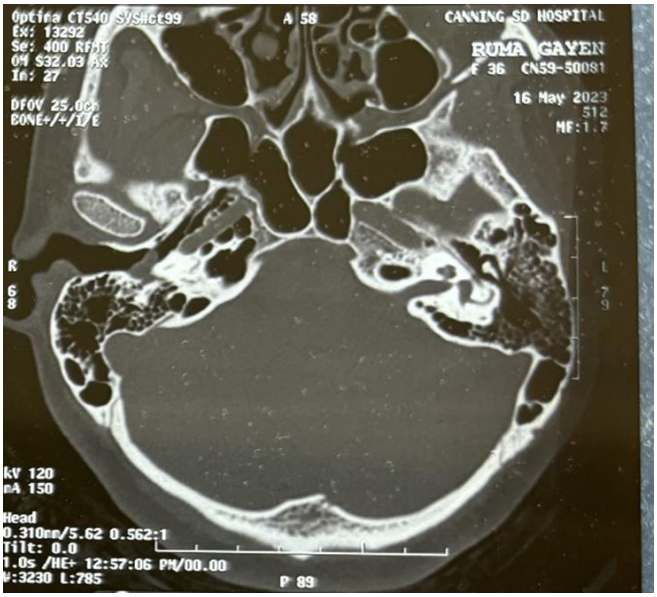

Fig. 2:- Axial CT scan of temporal bones showing a focal soft tissue density area in between the tympanic membrane and the promontory on the right side.

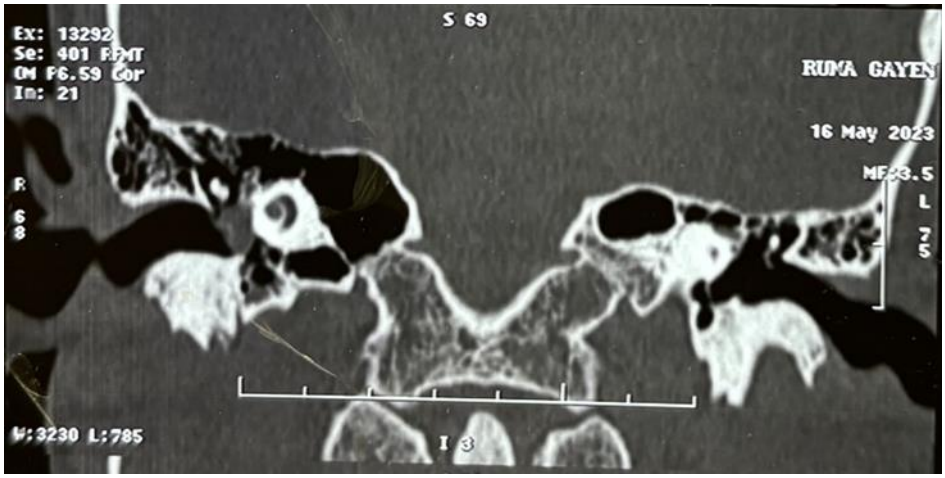

Fig. 3: Coronal CT scan of temporal bones showing a focal soft tissue dense area in between the tympanic membrane and the promontory on the right side.

The patient was operated under local anaesthesia. A post-auricular approach was adopted. Tympanomeatal flap was raised. The mass was bleeding moderately which was controlled by application of a cotton wool soaked in 1:1000 adrenaline solution. The mass was found to be occupying the mesotympanum only. The epitympanum and the posterior tympanum were found to be free. The mass was found to be in firm contact with the handle of the malleus and the long process of the incus. The mass was carefully dissected out to free it from the ossicles. The mass was adhered to the tympanic membrane. The tympanic membrane was torn at the area overlying the mass while the mass was dissected from it. The whole mass was removed in a single piece (Fig.- 4). The defect of the tympanic membrane was repaired with a temporalis fascia graft. The removed mass (Fig.- 5), which measured 0.8 cm x 0.6 cm was sent for histopathological examination.

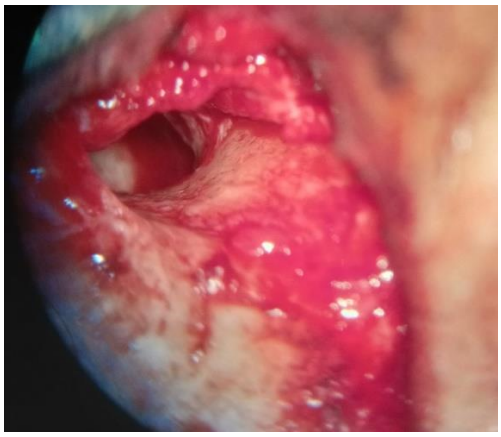

Fig. 4:- Intra-operative photograph of the right middle ear cavity after removal of the mass.

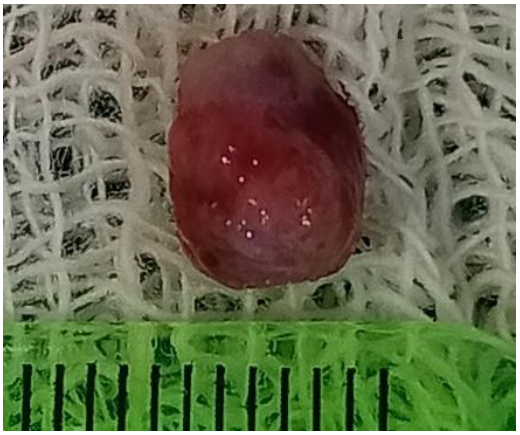

Fig. 5:- Post-operative photograph of the excised mass measuring 0.8 cm X 0.6 cm.

In the post-operative period, the patient recovered very well and after seven days she was discharged from the hospital.

The histo-pathological examination revealed 'Lobular Capillary Haemangioma' (Fig.- 6) comprising of lobules of capillaries, with no evidence of malignancy.

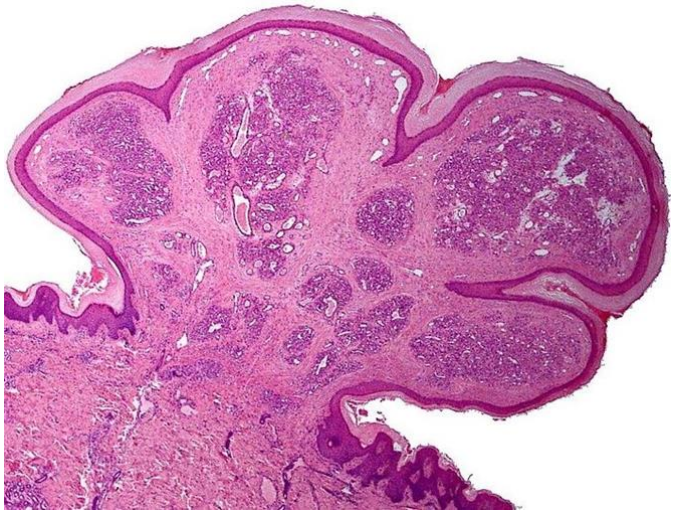

Fig. 6:- Histo-pathological picture of the specimen showing features of Lobular Capillary Haemangioma including lobules of capillaries.

The patient was followed-up for a period of 6 months. The graft was taken up satisfactorily and a very good functioning neo-tympanum could be seen after 3 months from the date of operation.

III. Discussion:

'Lobular Capillary Haemangioma' of the middle ear is a rare entity. There are multiple factors, such as trauma, hormonal influences, unknown angiogenic factor, inflammatory and infectious agents, which can lead to its development. The exact cause of it is not known. It is a benign tumour and presents mostly in neonates with an estimated incidence of 1 to 2.6 % at birth and 10 % in the first year of life. About 50% of these lesions regress within the first 5 years of life.[12]

In temporal bone haemangiomas, most common presenting manifestations are conductive deafness, pulsatile tinnitus, blood-stained ear discharge, earache or facial palsy. These features often lead to misdiagnosis as several common ear pathologies including glomus tumours (glomus tympanicum, glomus jugulare), aberrant carotid artery, high jugular bulb etc. , also present with similar clinical findings. In our case, the patient presented with the complaints of gradually progressive hearing loss, earache and perception of buzzing sound in the right ear. In the case reported by Pistario et. al., [7] the patient presented with a 10-year history of fullness of the ear, hearing loss and a history of ear bleeding in the previous year. A High-Resolution CT scan has been suggested as the primary investigation by most of the researchers. An MRI should also be done to delineate the nerves and vessels covering the mass. However, it should be kept in mind that MRI features of a haemangioma and a glomus are similar. Hence, histopathological examination should always be done to confirm the diagnosis.

Treatment of these lesions suggested in the literature is excision with clear surgical margins. The surgical technique depends on the tumour sizes, the degree of hearing loss, and the situation of the jugular bulb. Different approaches are often considered depending on the position of the lesion. Small lesions can be removed by transcanal approach. In cases of advanced lesions, transmastoid, translabyrinthine, middle cranial fossa or combined approaches may have to be considered. In our case, we preferred the transcanal approach to remove the lesion as it was limited within the middle ear cavity. Kostrzewa et. al. [13] described the utilization of CO_2 LASER assisted excision of tumour to reduce the danger of bleeding and for better visualization. Pavamani et. al. [12] reported the use of radiotherapy as the primary treatment of an inoperable capillary haemangioma involving the external and the middle ear, with a 5-year recurrence-free period. Although in recent years, propranolol is extensively used in treating haemangiomas of the other regions of the body, [14] its efficacy for intratemporal haemangiomas is uncertain.

IV. Conclusion:

'Lobular Capillary Haemangioma' of the middle ear is an extremely rare benign vascular neoplasm. It is very difficult to diagnose it clinically. Most often, histopathological examination is the only way to diagnose it. Complete surgical excision of the mass along with adequate follow up is the treatment of choice.

Conflict of interests: None.

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