

Intramuscular Capillary Haemangioma of the Temporalis Muscle: A Rare Case with A Review of the Literature

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ABSTRACT

An Intramuscular Haemangioma (IMH) is a benign mesenchymal tumour of the endothelial cells that accounts for less than 1% of all haemangiomas. Here we report the case of a capillary type intramuscular haemangioma in a five-year-old boy, only the fourth such case reported in literature, along with a relevant review of the literature. The lesion was surgically managed, with no recurrence in the follow up period till date.

Keywords: Male child, Muscle neoplasms/radiography, Temporal muscle/pathology

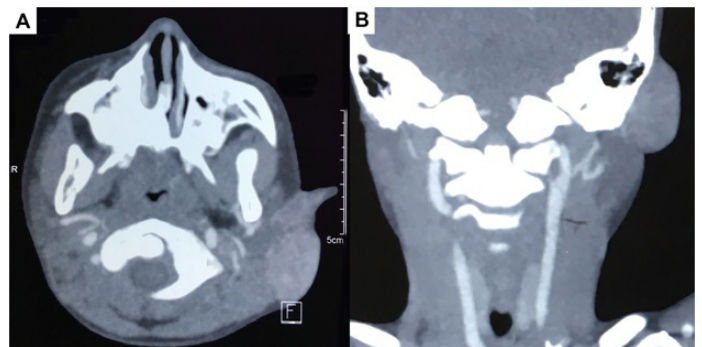
CASE REPORT

A five-year-old boy was brought to the Department of Otorhinolaryngology outpatient department with a painless swelling behind the left ear that had been gradually increasing in size for two years, with no other complaints.

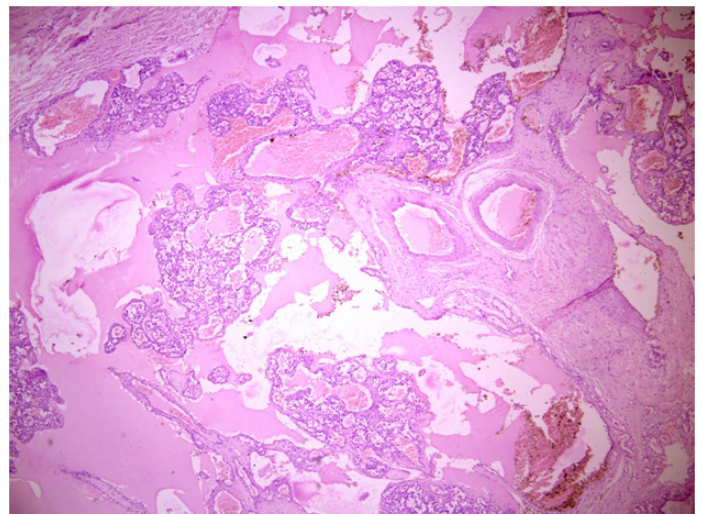
Physical examination revealed a single, soft, non-tender swelling, 4x3 cm in size, in the left post-aural region pushing the pinna forward, with visible dilated vessels on the surface of the swelling [Table/Fig-1]. The external auditory canal and tympanic membrane were normal.

Ultrasound examination showed a soft tissue mass with intense vascularity on colour doppler and an arterial waveform on spectral doppler. Fine Needle Aspiration Cytology (FNAC) of the lesion was non-diagnostic on account of a bloody specimen. Computed Tomography (CT) angiography revealed a vascular malformation of size 35 x 25 x 15 mm in the left post-aural region with feeders from the posterior auricular and occipital branches of left external carotid artery [Table/Fig-2].

The mass was excised en-bloc under General Anesthesia after ligation of feeder vessels, with an uneventful postoperative recovery. Histopathological analysis revealed scattered skeletal muscle interspersed with thin-walled vessels, consistent with intramuscular capillary haemangioma [Table/Fig-3]. The patient has been regularly following up for 16 months with no evidence of local recurrence.



[Table/Fig-2]: CT angiography revealed a vascular malformation of size 35 x 25 x 15 mm in the left post-aural region with feeders from the posterior auricular and occipital branches of left external carotid artery.



[Table/Fig-3]: Histopathological examination (H&E Stain) reveals scattered skeletal muscle interspersed with thin-walled vessels, consistent with intramuscular capillary haemangioma (10X magnification).



[Table/Fig-1]: A single swelling, 4x3 cm in size, in the left post-aural region pushing the pinna forward, with visible dilated vessels on the surface of the swelling.

DISCUSSION

An Intra Muscular Haemangioma (IMH) is a benign mesenchymal tumour of the endothelial cells. Accounting for only 0.8% of all haemangiomas. A total of 14% of IMH have been reported in the head and neck region, with the masseter (36%) and trapezius (24%) being the most commonly affected muscles [1]. IMH of the temporalis muscle is extremely rare with only 21 well documented cases in international literature, and only three reported cases of intramuscular capillary haemangioma [Table/Fig-4] [2-4].

Case	Author/Year	Age (years)/Sex	Treatment
1	Sharma et al., 1991 [2]	21/M	Surgery
2	Benateau et al., 1997 [3]	61/F	Surgery
3	Sharma et al., 2001 [4]	27/M	Surgery
4.	Present case	5/M	Surgery

[Table/Fig-4]: Details of reported cases of intramuscular capillary haemangioma in english literature [2-4].

Liston, in 1843, was the first to describe an IMH [5]. In 1972, Allen and Enzinger classified them on the basis of vessel size into capillary, cavernous and mixed types, with capillary variant being the most commonly encountered [6].

A number of theories exist to explain the aetiology of the tumour; however, none are accepted universally till date. It has been hypothesized that these tumours may be of congenital origin due to the high incidence in the early years of life, with trauma and hormonal changes considered as important additional factors for the proliferation of embryonic vascular tissue [1].

IMH often present as a slow growing mass with distinct margins, without pulsations or discolouration of the overlying skin. Due to their rarity and absence of the above-mentioned vascular signs, it is difficult to diagnose IMH without radiological investigations. Due to its presentation as a soft to firm non-tender mobile mass, a number of pathologies may be considered in the differential diagnosis, including enlarged lymph nodes, neurofibroma, lipoma, dermoid cyst, soft-tissue sarcoma, temporal arteritis, and myositis ossificans [1,7]. IMH may be readily differentiated from these pathologies with the help of different modes of imaging. Computed tomography helps to define the size and relations of the tumour. Magnetic Resonance Imaging (MRI) is more useful in defining its vascular nature, with the tumour appearing hyperintense on T2-weighted imaging due to volume of stagnant blood that is easily distinguished from surrounding muscle [8]. Angiography helps to detect feeder vessels, and is useful for planning preoperative embolization.

To the best of our knowledge, only 25 cases of intramuscular haemangioma of temporalis have been reported in English literature, only 21 of which are well documented [7]. Majority of these cases (85.6%) were either mixed or cavernous haemangiomas, with only three cases of capillary type (14.4%). A slight female predominance was seen (sex ratio of 0.82), with a mean age of 33.7 years [7].

Generally, surgical excision along with a rim of normal tissue was the mainstay of treatment in majority of the cases, with only one case kept under long-term follow-up without surgical intervention. Use of corticosteroids, sclerosing agents and radiotherapy have also been proposed in the past as an alternative or adjunct to surgical excision for reduction in tumour bulk [9].

Local recurrences have been reported, and are more common with the mixed (28%) and capillary varieties (20%); only 9% of cavernous types recurred [10].

CONCLUSION

Intramuscular haemangioma of the temporal region, although rare, is an important differential diagnosis that must be kept in mind when dealing with a post-aural swelling. The importance of identifying them lies in their ease of excision once the feeding vessels are identified and ligated, and the possibility of local recurrence is long term, especially in the capillary or mixed varieties. Thus, a meticulous long-term clinical and radiological follow-up is necessary in cases of IMH.

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