Haemangioma of the Left Lateral Surface of Tongue, In an Adult Patient: A Rare Case Report

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ABSTRACT

Introduction: Haemangiomas are rapidly growing vascular abnormalities that are benign in nature and often manifest in the neonatal period or during childhood. It is most commonly seen in the head and neck region (60-70%). It is rarely found in the oral cavity and occurrence in the tongue is even rarer. Haemangiomas in the oral cavity are always of clinical importance and require an appropriate treatment.

Case report: A 26-year-old male patient reported to our unit with history of swelling on the left lateral surface of tongue and difficulty in swallowing and breathing since 3 months. The patient was diagnosed to have vascular lesion after FNAC and MRI report. Patient was treated by embolisation of left lingual artery followed by surgical excision. Post op biopsy report showed the features of cavernous haemangioma.

Conclusion: Haemangioma continues to be used as a clinical and pathological description for many different types of vascular anomalies. The treatment modality should be planned according to the diagnosis and prognosis of the haemangioma.

Keywords: Haemangioma, swelling on surface of tongue, vascular abnormalities

INTRODUCTION

Haemangioma (Greek: Haima-blood; angeion-vessel, oma-tumor) by definition can be defined as a tumor of dilated blood vessels.[1] Haemangiomas are rapidly growing vascular abnormalities that are benign in nature and often manifest in the neonatal period or during childhood.[2,3] They contribute to 7% of all benign tumors and 60-70% of these occur in the head and neck region[4], with a female predilection of 3:1. It is rarely found in the oral cavity and occurrence in the tongue is even rarer. These are identified by rapid endothelial cell proliferation in early infancy, followed by involution over time.[4] Most true haemangiomas involute with time, but 10-20 % of them incompletely involute and require post adolescent ablative treatment. We report a case of haemangioma of the tongue in an adult male patient.

CASE REPORT

A 26-year-old male patient reported to the Department of Oral and Maxillofacial Surgery, Army college of Dental sciences, Secunderabad India, with a chief complaint of swelling on the left side of tongue and difficulty in swallowing and breathing since 3 months. The patient's history revealed that the swelling was of peanut size when it was first noticed around 3 months back and it gradually increased to attain the present size. There was no complaint of pain, difficulty in phonation, and altered taste sensation, and he did not give any history of trauma, bleeding or ulceration of the tongue.

History excluded any similar condition reported in any of his family members. Patient's general physical and extraoral examination was normal. On intraoral examination, there was a solitary oval-shaped swelling measuring 5 ×6 cm in size on the left postero-lateral border of the tongue partly involving dorsum and the ventral surfaces. The surface of the swelling was pinkish in colour with well defined borders. There was no ulceration or bleeding or any discharge from the tongue, and surrounding areas of the tongue were normal. Floor of the mouth was not raised. On palpation, all inspectory findings were confirmed, the lesion was soft in consistency, non-tender, compressible, non-fluctuant, no fluid thrill was present and it was not fixed to the underlying structures (Fig.1) The lesion blanched on compressing and filled again on releasing. Based on the history and clinical findings, a clinical diagnosis of vascular malformation/Haemangioma was considered. Differential diagnosis included lymphangioma, lingual varix and mucocele.

FNAC was performed which yielded about 2ml of frank blood. H and E stained smears showed blood elements, without any epithelial or stromal cells. All blood investigations were within the normal limits. MRI revealed a well defined predominantly T_2 hyperintensity lesion of size approximately 55x41x42 mm, (Fig.2) involving left half and posterior aspect of tongue with normal contour. Mid line shift to right because of bulging. FNAC and MRI reports confirmed the findings of a vascular lesion. Before the surgical procedure, Angiogram was performed to identify the feeding vessel and embolise the feeding vessel on the same appointment. Angiogram was carried out through the right femoral artery and both internal and external carotids were cannulated selectively. Angiogram runs were taken**.** Right carotid angiogram was normal whereas left external carotid angiogram showed a large tumour of the tongue feeded by the left lingual artery. Left lingual artery was then selectively canulat-

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ed and embolised with gelfoam. Surgical excision of lesion was planned after 24 hours of embolization under general anaesthesia. Submandibular incision with lip split was used to access the lesion. Mandibular split osteotomy was carried out to visualise the extent of tumorAn incision was given over the lesion to completely tease out the tumor from its bed (fig.3). Osteotomy cuts were approximated and fixed using Titanium plates and screws. Closure was done using 3-0 vicryl intra-orally and 3-0 silk extra-orally. Post operative excisional biopsy confirmed the feature of cavernous haemangioma (Fig.4).

DISCUSSION

Hemangiomas are the most common benign tumours of the head and neck in children, but their occurrence on the tongue is extremely rare. The tongue requires special consideration because of its susceptibility to minor trauma and consequent bleeding and ulceration, swallowing difficulties, and breathing problem, although the major concern is cosmetic in most cases. The hemangiomas appear as soft mass, smooth or lobulated, and sessile or pedunculated and may vary in size from a few millimeters to several centimetres.[6,7] The color may vary from pink to red purple and blanching on the application of pressure is evident.[8] In our case the lesion was pink in color.

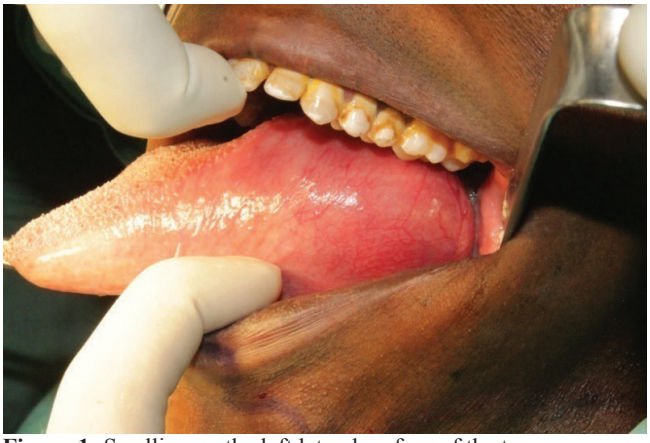

Figure-1: Swelling on the left lateral surface of the tongue

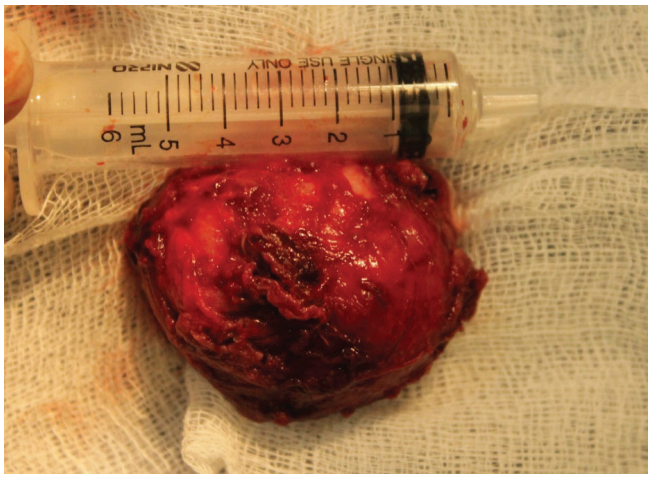

Figure-3: Lesion after surgical excision

Hemangiomas are characterized by 3 stages: Endothelial cell proliferation, rapid growth and spontaneous involution. Monocytes are considered the potential ancestors of hemangioma endothelial cells. Vascular endothelial growth factor (VEGF), basic fibroblast growth factor (BFGF) and indole-amine 2, 3-dioxygenase (IDO), which are found in large amount during proliferative stages, are believed to be the contributing factors for growth.[2]

Imaging plays an important role in diagnosis, which includes Angiography, Ultrasonography, Contrast enhanced MRI, CT scans and Doppler ultrasonography, etc.[8] Ultrasound and MR imaging are both reasonable diagnostic tools for a suspected hemangioma.[11] As a non-invasive, inexpensive,and increasingly available imaging modality, ultrasonography is ideal for evaluating a suspected hemangioma that is small in size and superficial in location. MR imaging is helpful in evaluating deep or large soft tissue masses. MR imaging shows a well-marginated soft tissue mass.[11] T1-weighted sequences show a homogeneouslesion with intermediate signal intensityduring the proliferative phase, and a heterogeneous lesion with small focal areas of fat replacement during the involutional phase.[12.13] On T2-Weighted images, lesions ap-

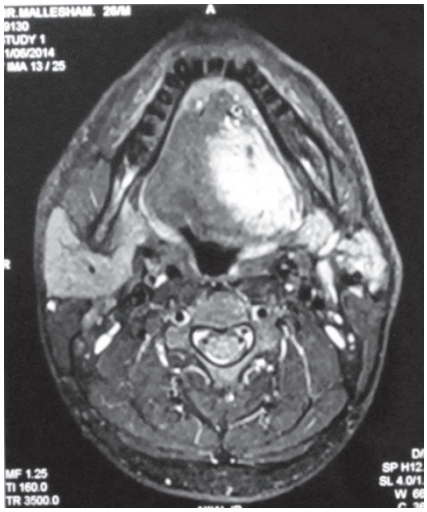

Figure-2: MRI of tongue showing the extent of lesion

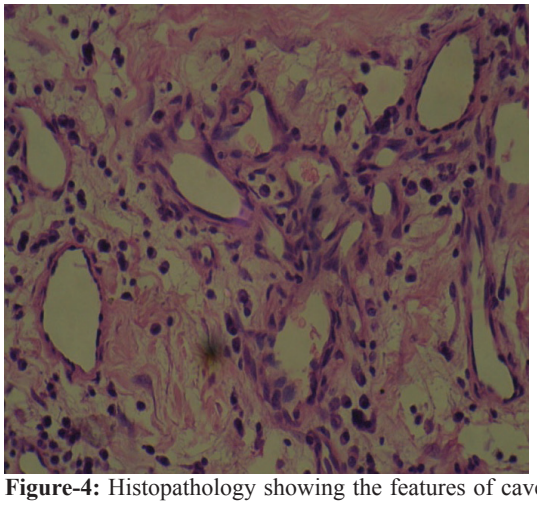

Figure-4: Histopathology showing the features of cavernous haemangioma

pear homogeneous and moderately hyperintense during the proliferative phase and more heterogeneous while involuting.Particularly deep or large lesions may require MR imaging for complete characterization.[11] In our case the lesion was T_2 hyperintensity.

The histological features are dependent on the stage of the lesion. In the proliferative phase, the lesion is highly cellular and contains plump proliferating endothelial cells and pericytes. Vascular channels are not prominent whereas, in the Involutive phase**,** the endothelial cells are flattened, the cell turnover is normal and vascular channels filled with blood cells predominate, and the lesion is eventually replaced by fibro fatty tissue.[14]

In our case post operative excisional biopsy revealed a cavernous haemangioma of the tongue which showed the presence of large dilated blood sinuses, with thin luminal walls with endothelial lining.

As far as the management of these lesions is concerned, useful approach to the management of haemangiomas can be based on the stage of the lesion (proliferative or involutive phase), type of lesion (superficial, deep, compound) and the management of residual deformity.[14]

Various treatment modalities are present that include wait and watch policy, for spontaneous involution, intralesional and systemic corticosteroid treatment, embolization, excision, electrolysis and thermocautery, immunomodulatory therapy with interferon alfa-2a, and laser photocoagulation.[9] Laser energy to photocoagulate vascular lesions is also an area of interest. Currently, sclerotherapy is employed largely because of its efficiency and ability to conserve the surrounding tissues.[10] Resection remains the mainstay of treatment for deep haemangiomas.[12] In our case lesion was deep and it was obstructing the airway therefore we planned the surgical excision of lesion.

CONCLUSION

Haemangioma continues to be used as a clinical and pathological description for many different types of vascular anomalies. Its occurrence on the posterior lateral surface of tongue is rare. Early detection and biopsy are crucial in determining the clinical behavior of the tumor and potential complications. The treatment modality should be planned according to the diagnosis and prognosis of the haemangioma.

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