Journal of Applied Dental and Medical Sciences
NLM ID: 101671413 ISSN:2454-2288
Volume 6 Issue 1 Jan-March 2020

Case Report

Cavernous hemangioma of the tongue: A rare case report

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ARTICLE INFO

Keywords:
Pulpectomy, Deciduous Teeth, Necrosis Pulp, Apical Periodontitis, Ledermix

ABSTRACT

Hemangiomas are benign tumor of dilated blood vessels. It is rarely seen in oral cavity and when present can cause considerable morbidity to the patient, therefore it requires prompt intervention. The present case report deals with hemangioma of tongue in a 14 yr old boy which was removed by surgical excision.

INTRODUCTION

Vascular lesions of the maxillofacial region are classified as either: (i) hemangiomas or (ii) vascular malformations/arterio-venous malformations[1]. Hemangiomas are the most common cutaneous tumor of infancy and demonstrate rapid growth followed by a slow spontaneous involution or regression within five to seven years. Unlike hemangiomas, vascular malformations enlarge proportionately with the growth of the child and do not undergo spontaneous involution[1]. Hemangiomas are the most common tumors in infants, whereas arteriovenous malformations (AVMs) are the least common, comprising about 1.5% of the total reported cases[2]
Hemangioma is histologically further classified into capillary and cavernous forms. Capillary hemangioma is composed of many small capillaries lines by a single layer of endothelial cells supported in a connective tissue stroma of varying density, while cavernous hemangioma is formed by large, thin walled vessels, or sinusoids lined by epithelial cells separated by thin layer of connective tissue septa.[3]
Hemangiomas are considered as benign tumors, being characterized by 3 stages: Endothelial cell proliferation, rapid growth and at last spontaneous involution. The pathophysiology of hemangiomas is attributed to genetic and cellular factors, mainly to monocytes, which are considered the potential ancestors of hemangioma endothelial cells. Imbalance in the angiogenesis, which causes an uncontrolled proliferation of vascular elements, associated with substances such as 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 cause[4]

Case report

A 14 yr old male patient has come to our centre with a chief complaint of swelling on his tongue. On examination it was found to be a lobulated mass of about 1 × 1.5 cm on the left lateral border of tongue. It was smooth and has a bluish hue. Their was no associated history of pain or any other discomfort . On palpation the swelling was soft to firm in consistency, non mobile, non tender, afebrile with no palpable thrills and blanching on compression. The swelling appeared 2-3 yrs back according to the patient and is been increasing in size ever since. The provisional diagnosis of hemangioma was made based on the appearance and progression of the lesion along with differential diagnosis of a granular cell myoblastoma, angiomyolipoma, angiosarcoma, hemangiosarcoma and Kaposi's sarcoma.
The routine blood and urine investigations were normal. Color Doppler ultrasound revealed hypoechoic, circumscribed well defined lesion measuring 1 × 0.4 cm seen in the left anterior lateral aspect of the tongue with arterial flow. Surgical excision was carried out under local anesthesia. During the surgical procedure, a thread was tied around the base, and the lesion was stretched in an upward direction in order to get maximum accessibility. The mass was then excised out, and interrupted sutures were placed (Figures 3). During the surgical procedure,

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minimal amount of bleeding from the site was observed. The specimen was then sent for a histopathological examination. The healing was uneventful after a period of 1 week and complete healing was seen after 1 month. Histopathological examination confirmed the definite diagnosis of cavernous hemangioma. Patient follow up was done for 6 months and all the tongue movements were normal and there was no recurrence of the similar lesion.

Figure 1 & 2 : Preoperative picture of lesion

Fig 3 : Site after suturing

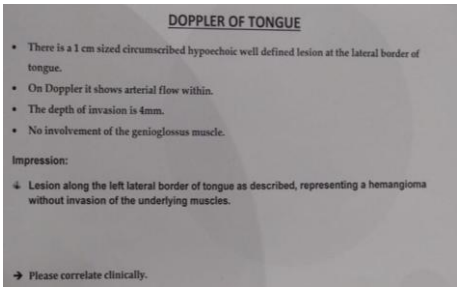

Fig 4: Colour Doppler report

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 hemangioma appears as soft mass, smooth or lobulated, and sessile or pedunculated and may vary in size from a few millimeters to several centimeters [5,6]. They are usually deep red and may blanch on the application of pressure and if large in size, it might interfere with mastication [7,8]. The superficial hemangiomas are often lobulated, and blanch under finger pressure and the deeper lesions tend to be dome-shaped with normal or blue surface coloration, and they seldom blanch. A lesion with a thrill or bruit or with an obviously warmer surface, is most likely a special vascular malformation, called arteriovenous hemangioma (arteriovenous aneurysm, A-V shunt, arteriovenous malformation), with direct flow of blood from the venous to the arterial system, bypassing the capillary beds.

Various syndromes that are associated with the vascular malformation include the Osler-Weber-Rendu syndrome, Sturge-Weber syndrome, and blue rubber bleb nevus syndrome. A differential diagnosis of granuloma fasciale, insect bite, pyogenic granuloma, and angiosarcoma can be given for this condition.

Clinical diagnosis was based on histopathological evaluation, which was confirmed to be a cavernous hemangioma. Histologically, the appearance of these lesions depends on the stage of the evolution. Early lesions may be very cellular with solid nests of plump endothelial cells and little vascular lumen. Established lesions comprise of well-developed, flattened, and endothelium-lined capillary channels of varying sizes in alobular configuration. Involuting lesions show increased fibrosis and hyalinization of capillary walls with luminal occlusion [9]. In cavernous hemangioma, there is presence of large dilated blood sinuses with thin walls each showing an endothelial lining. The sinusoidal spaces are usually filled with blood although there might be presence of lymphatic vessels [10].

Although hemangioma is considered one of the most common soft tissue tumors of the head and neck, it is relatively rare in the oral cavity and uncommonly encountered by the clinicians. Radiographic imaging is indicated preoperatively in selected cases where large lesions may impinge on vital anatomical structures, such as the facial nerve or orbit. Computed tomography (CT) and magnetic resonance imaging (MRI) can also be used for volumetric analysis of hemangiomas and vascular malformations. Imaging resources can also be useful in both diagnostic differentiation and analysis of lesion features with regard to its size, extension and location, as well as for follow up of lesions treated under a systemic therapy. In the present case as the biopsy and fine needle aspiration cytology was contraindicated, we used Color Doppler ultrasound to identify the feeding vessel, which helped to legate during surgical procedure. Baba and Kato reported a case of hemangioma with pleboliths in the floor of the mouth and Doppler ultrasonography, CT, MRI was used to diagnose the lesion[11]

Management of hemangioma depends on a variety of factors, and most true hemangioma requires no intervention. However, 10-20% requires treatment because of the size, exact location, stages of growth or regeneration. There are many treatment modalities reported in the literature for head and neck hemangiomas, including 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.Recent interest has centered on interstitial delivery of laser energy to photocoagulate vascular lesions. Currently, sclerotherapy is employed largely because of its efficiency and ability to conserve the surrounding tissues.

Among the sclerosing agents available, excellent results have been

reported for sodium morrhuate, sodium sulfate tetradecyl, polidocanol and ethanolamine oleate, and hypertonic glucose solution[11] .Growing hemangioma can be treated effectively by systemic drug therapy, sclerotherapy, laser therapy or combined therapy. Transcutaneous and contact applications of laser energy have been studied with the argon and Nd: YAG lasers while the 585-nm flashlamp - pulsed dye laser can be used in cutaneous and subcutaneous hemangiomas. Vesnaver applied photocoagulation with interstitial Nd: YAG laser in 111 patients with vascular lesions in the head and neck region and he concluded that, Nd: YAG laser is a safe and effective tool for treating vascular lesions.

Hemangiomas of the tongue are rare lesions which can cause distressing problem to the patients, producing cosmetic deformity, recurrent hemorrhage, and functional problems with speaking, mastication and deglutition. The treatment depends on lesion location, size and evolution stage, and the patient's age. Due to its side effects, radiotherapy and chemotherapy would not be suitable as a treatment choice for tongue lesion. Swallowing, chewing and speaking function disabilities were seen in the cases where CO2 laser was applied. Surgery is usually indicated when there is no response to systemic treatments, or even for aesthetic reasons, being performed as a simple excision in combination or not with plastic surgery. Conservative or further aggressive forms of treatment may be tried for the hemangiomas of the tongue. Both treatment methods have disadvantages. In the conservative treatment, recurrences may be frequent. On the other hand, aggressive treatment could also cause function loss. However, the results of cryotherapy have been reported to have high success rates. Kutluhan [12] used plasma knife surgery for excision of hemangioma of tongue.

Conclusion

The appearance of cavernous hemangioma is a rare occurrence on the tongue. Early detection and biopsy are crucial in determining the

clinical behavior of the tumour and potential complications. Surgical excision is the method of choice in isolated small sized hemangiomas.

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