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Case Report

Dentistry Section

Cavernous Hemangioma of Tongue: Management of Two Cases

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ABSTRACT

Hemangiomas are benign tumours of infancy and childhood, most commonly located in the head and neck region and occur more frequently in the lips, tongue and palate. The treatment depends upon lesion location, size and evolution stage and the patient's age. This paper describes the management of cavernous hemangioma in a 2 -year -old child and 14 -year -old child using different approaches.

Keywords: Corticosteroids, Electrocautery, Hemangioma, Surgery

CASE REPORT

Case 1

A 14-year-old male child was referred to the Department of Peadiatric dentistry for the evaluation of a bluish swelling on the lateral surface of anterior two thirds of the tongue on left side. History revealed that the swelling was present since birth. There was no history of trauma. It was occasionally associated with pain and bleeding. On general examination the patient was normally built for his age with no defect in stature or gait. No relevant medical history was observed. On intraoral examination, there was a growth measuring about 3cm x 2cm, which was blue in colour on the left lateral surface of the tongue which appeared to be sessile with no underlying attachment or relation with the muscles [Table/Fig-1]. The borders were well defined and there was no ulceration seen on the surface of the lesion. The growth was soft to palpate, and it showed blanching on application of pressure. A provisional diagnosis of hemangioma of tongue was given based on the clinical findings. Since the lesion was a circumscribed lesion hence ultrasonography was not done in case 1.

After completion of pre anaesthetic checkup patient was posted for general anesthesia. Initially stay sutures were given to the tongue to prevent fall back during procedure. Kole's incision marking was done. Resection of the pathological entity was done [Table/Fig-2]. Post resection was done with cautery to prevent further bleeding [Table/Fig-3]. After resection of pathological entity, it was sent for histopathological examination followed by suturing of the tongue [Table/Fig-4]. The healing was uneventful after a period of one week and complete healing was seen after one month, also patient recalled after five months for review [Table/Fig-5]. The histopathology report confirmed the diagnosis of cavernous hemangioma with void capillary vessel.

Case 2

A 2-year and 3-month old boy presented with increased tongue size and eruptions on tongue surface since six months [Table/Fig-6]. Tongue appeared larger at birth and had increased to the present size. Papillary eruptions were present on the anterior two thirds of the tongue associated with burning sensation which was aggravated on consumption of spicy and hot beverages. His parents also complained of difficulty in speech. A provisional diagnosis of hemangioma of tongue was given based on the clinical findings. The two noninvasive imaging techniques that are most useful in the examination of vascular malformations are MRI and sonograph. Due to economic constraints of the patient we planned soft tissue ultrasonography. Ultrasonography of the tongue demonstrated mixed echogenous lesion (22 x 13 mm) with small cystic areas and increased vascularity confirming the diagnosis of hemangioma causing macroglossia [Table/Fig-7]. Treatment was started with Omnacortil syrup (prednisolone sodium) 5 ml thrice daily for two weeks and then the dose was tapered for another two weeks. After two weeks there was complete resolution of the papillary eruptions, patient recalled after three months for evaluation [Table/Fig- 8].

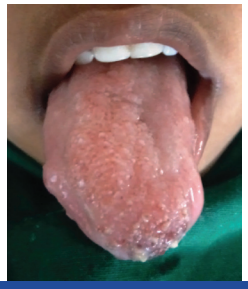
DISCUSSION

The term hemangioma has been commonly misused to describe a large number of vasoformative tumours. These lesions represent the most common growths in infancy and childhood, and may vary from small innocent birthmarks to large disfiguring tumours [1]. Hemangiomas usually appear 2-4 weeks after birth; grow rapidly till the age of 6 – 8 months and then slowly develop. By age 5-8 years, they start to involute and spontaneously regress in 70% of the cases [2].

Historically, hemangiomas have been classified in a variety of ways. According to Enzinger and Weiss, hemangiomas are broadly




[Table/Fig-1]: Hemangioma on ventral surface of tongue **[Table/Fig-2]:** Surgical resection of the hemangioma **[Table/Fig-3]:** Cauterization of resected area **[Table/Fig-4]:** Suturing technique


[Table/Fig-5]: Six months post op **[Table/Fig-6]:** Macroglossia with eruptions on tongue -CASE -2 **[Table/Fig-7]:** Ultrasonography of the tongue demonstrated mixed echogenous lesion with small cystic areas and increased vascularity – CASE - 2 **[Table/Fig-8]:** Three months post op-CASE -2

classified into capillary, cavernous, and miscellaneous forms like verrucous, venous, arteriovenous haemangiomas, and so forth [3]. An important descriptive classification is related to the depth of soft tissue involvement' superficial, deep and mixed [4]. Hemangiomas are considered benign tumours of infancy and childhood and have a different life cycle being characterized by three stages endothelial cell proliferation, rapid growth, and at last spontaneous involution. They are characterized by hyperplasia of blood vessels, usually veins and capillaries in a focal area of submucosal connective tissue. It is almost never encapsulated [5].

Whether this condition is a neoplastic or reactive state is uncertain; a reactive cause is favoured. Few of the reactive causes are namely hormonal changes, infections and trauma.

The majority of hemangiomas involve the head and neck. However, they are rare in the oral cavity but may occur on tongue, lips, buccal mucosa, gingiva, palatal mucosa, salivary glands, alveolar ridge, and jaw bones [6]. Among the different sites of head and neck hemangiomas, tongue requires special consideration in view of it being a mobile inquisitive organ resulting in increased susceptibility to minor trauma and consequent bleeding and ulceration, swallowing difficulties, breathing and aesthetic problems.

Hemangiomas show a notorious higher prevalence in females (3:1 to 7:1 predilection) and the superficial type is the most frequent one, unlike the present cases where both the hemangiomas were seen in male children [3]. In our case, no similar lesion was found in other body systems and the tongue was the only site for the hemangioma; whereas 20% of haemangiomas are present at more than one site [6].

A provisional diagnosis of hemangioma and a differential diagnosis of arteriovenous malformation were given in the first case. Hemangiomas are often circumscribed lesions which rarely affect bone and are most commonly present on tongue, lips and buccal mucosa. The lesion in the first case was a well circumscribed lesion on the ventral surface of the tongue. On the other hand AV malformations are poorly circumscribed lesions which may affect bone. AV malformations are also difficult to resect resulting in surgical hemorrhage but there was no extensive hemorrhage during the resection of the lesion in the first case. Finally based on the histopathological report a diagnosis of cavernous hemangioma was made.

The treatment plan established for haemangiomas must consider aspects such as size, location, lesion hemodynamics, patient's age and viability of the technique to be used. Systemic corticosteroids have been established as the most efficient medical therapy for common cutaneous infantile hemangiomas if started early in the proliferative phase. Oral prednisolone has been used for 30 y and remains the first line therapy for difficult hemangiomas. The dosage commonly used is 3-5mg/kg per day of oral prednisolone between the first & thirtieth months. The main side effects of high dose therapy in long term are cushingoid features, influence on growth, and susceptibility to serious infections, appetite changes, behavior changes, polyuria, thrush and gastrointestinal discomfort [7].

When there is no response to the systemic treatment or if there is an aesthetic complaint, surgery is indicated. There are many reports on using preoperative embolization or applied sclera therapy or radiotherapy before or concomitant with surgery. However, embolization technique has two major disadvantages; one is the risk of embolization material reaching cranial cavity via external and internal carotid arteries hence the need for an experienced radiologist to perform this procedure and the temporary blockage of flow [8]. Sclerotherapy is successfully utilized in the treatment of extraoral lesions. Among the sclerosing agents available, excellent results have been obtained for sodium morrhuate, sodium sulfate tetradecyl, polydecanol and ethanolamine oleate and hypertonic glucose solution. However, pressurized bandage cannot be applied to the region after injection of sclerotic agent in intraoral lesions. Moreover use of sclerotherapy requires multiple visits [9]. Radiotherapy is useful to reduce the tumoral volume in intraosseous hemangiomas. Nevertheless, it has a lot of adverse effects such as damage to the normal adjacent tissues growth, residual scarring and malignancy. So, radiotherapy is considered an unacceptable therapeutic option [10]. In our case it seemed that with a judicious surgical approach, there was no need for adjunctive measures as surgical excision of the lesion was followed by cautery to reduce postoperative bleeding.

Propranolol, a nonselective beta-blocker, has recently been introduced as a novel modality for the treatment of proliferating haemangiomas by Le´aute´-Labre`ze et al., [11]. The exact mechanism of action of propranolol in the treatment of haemangiomas remains unclear, but vasoconstriction, down-regulation of angiogenic factors such as VEGF and bFGF and up-regulation of apoptosis of capillary endothelial cells may be responsible for the reduction of haemangiomas. 2–3 mg/kg/day propranolol should be administered, divided in 2–4 doses per day [12]. Potential side effects of beta-blockers include bradycardia, hypotension, hypoglycemia; rash, gastrointestinal discomfort/reflux, fatigue and bronchospasm, all are rare and observed at higher doses (>2 mg/kg/day).

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