Oral haemangiomas- Series of two case reports and review of management

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Abstract

Haemangiomas are one of the most common of all human birth defects and are based on vascular tissues. These lesions are mainly identified into two groups which are named as (a) capillary and (b) cavernous haemangioma. Capillary haemangioma consists of small capillary vessels which show lobules formation. Cavernous haemangioma consists of large dilated vessels and they can reach large sizes. Many treatment modalities are evaluated in which some modalities are successful and some are quite disappointing. Surgical excision, irradiation, CO₂ freezing, sclerosing agents, cauterezation, steroid therapy and watchful waiting are among the treatment methods evaluated. 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. In the present series of two case reports, the management of haemangioma was done using two different modalities depending on their presentation so that major complications can be avoided and more attention should be paid to more conservative treatment modalities.

Keywords: Haemangiomas, treatment modalities, tumors

Introduction

According to Mulliken and Glowacki in 1982, vascular anomalies are categorised into two major groups: (a) Haemangioma and (b) Vascular malformations. Haemangioma is the most frequently encountered vascular soft-tissue abnormality. It is estimated that haemangiomas comprise 7% of all benign soft-tissue tumours. More than 50% of these lesions occur in the head and neck region. They show a higher prevalence in females than males (3:1 ratio). Haemangiomas are present shortly after birth. At this stage, it may be confused with other red lesions of birth, but rapid proliferation and vertical growth trigger the diagnosis. They follow a predictable course with three distinct developmental phases: proliferation, quiescence and involution. In most haemangiomas, 80% of the proliferation occurs by 3 months of life but may last longer. During proliferation, rapid growth can lead to exhaustion of blood supply with resulting ischaemia, necrosis, ulceration and bleeding. Haemangiomas are further sub-classified based on their histological appearance as (a) capillary haemangioma and (b) cavernous haemangioma. Capillary haemangioma consists of small capillary bodies that organise lobularly. Cavernous haemangioma consists of wide and dilated vessels and can reach large sizes. Intra-orally, the possible sites of occurrence are lips, tongue, buccal mucosa, gingiva and palate. The lesions on the cheeks and tongue can be traumatised while chewing and bleeding can occur. Also, some patients with haemangioma on their tongues have complained about burning and pain sensations. Haemangioma possesses an extremely challenging treatment dilemma for surgeons and patients. Most of the haemangiomas involute but if they do not involute, they impair the vital functions and need proper intervention by oral physicians.

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Case 1
A 37-year-old female patient reported to the Department of Oral Medicine and Radiology with the chief complaint of growth in relation to the left lateral surface of the tongue for the last 3 months. The patient was asymptomatic and unaware of any growth on the tongue 3 months ago after which she noticed growth in relation to the left lateral side of the tongue which was slow-growing, painless, associated with bleeding and caused difficulty in chewing food from the left side. There was also a history of burning sensation concerning the growth on taking spicy food. The symptoms got relief after taking medications (OTC) and pressure packs. On extra-oral examination, there was no appreciable facial asymmetry evident from the frontal profile. No abnormality was detected with the cervical lymph node and neck regions. On intraoral examination, a solitary, localised, well-defined, pedunculated, exophytic growth measuring approximately 3 cm × 2 cm in diameter was evident in relation to the left posterior lateral border of the tongue, which was dark-red to purplish-black covered with corrugated and friable superficial mucosa and was traumatised [Figure 1]. On palpation, the growth was non-tender, soft in consistency, non-pulsatile, non-reducible which bled on slight provocation. A diascopy test was performed and it was negative. Since the lesion was circumscribed, ultrasonography was not done. A provisional diagnosis of pyogenic granuloma in relation to the left posterolateral border of the tongue was given with a differential diagnosis of traumatic fibroma, papilloma and haemangioma. Total excision biopsy was carried out under local anaesthesia and the specimen was sent for histopathological evaluation. The sub-epithelium showed proliferated capillaries and dilated vascular channels filled with blood and confirmed the diagnosis of capillary haemangioma [Figure 2]. A follow-up was taken after 5 days and the lesion was healed [Figure 3].

Case 2
A 32-year-old male patient reported to the Department of Oral Medicine and Radiology with the chief complaint of swelling on the right upper front region of the jaw over the labial vestibule and gingiva for 5 years. History dates to 5 years when the patient experienced swelling on the right upper front region of the jaw over the labial vestibule and gingiva which was small initially. It gradually increased in size and attained its present size. Since then, sometimes, the swelling subsides partially after the watery discharge, and then, after some time, regains the original size. The swelling has never been painful or associated with pus or blood discharge. On extra-oral examination, the face was bilaterally symmetrical. On intraoral examination, there was the presence of an irregular oval-shaped swelling on the right labial mucosa and gingiva with the 12, 13, 14 tooth region [Figure 4]. It had a lobulated appearance with a bluish surface discoloration and having an irregular surface with no discharge. There were no visible pulsations. It had a corrugated appearance with a bluish discoloration and a punctum. On palpation, inspectory findings were confirmed. The lesion was soft, well-circumscribed, non-tender and compressible having a slightly corrugated surface. It blanched under finger pressure. The surrounding mucosa was normal and the swelling was not fixed to the underlying structures. It was slightly reducible and fluctuant but no pulsations were palpable. The diascopy test was positive. A provisional diagnosis of haemangioma with the right upper labial vestibule was given. IOPA in relation to the 12, 13 and 14 revealed no significant findings [Figure 5]. A contrast-enhanced CT was done which revealed a well-defined isodense to hypodense lesion measuring 17.9 mm × 7 mm along the gingival surface of the alveolar arch of the maxilla on the right side. It also showed a homogenous enhancement causing mild pressure scalloping of the underlying bone with no resorption [Figure 6]. CECT findings were suggestive of haemangioma. The patient was given an intraslesional injection of 1 mL of the sclerosing agent: 3% setrol (sodium tetradecyl sulphate) mixed with lignocaine HCL once a week for 3 weeks [Figure 7]. A tuberculin syringe was...
Discussion

The term haemangioma was originally used to describe any vascular tumour-like structure, whether it was present at or around birth or appeared later in life. The term is comprised of the Greek words ‘haema’ which means blood, ‘angio’ meaning vessel and ‘oma’ meaning tumour. Histologically, haemangiomas are composed of hyperplastic endothelial cells, which line the inner surface of the blood vessels in the human body, with the capacity for intensive proliferation. The diameter of the blood vessels is important in the classification of haemangioma to capillary and cavernous types. The capillary type, also known as the strawberry haemangioma, is composed of small thin-walled vessels of capillary size that are lined by a single layer of flattened or plump endothelial cells and surrounded by a discontinuous layer of pericytes and reticular fibres. The cavernous type is characterised by large blood-filled spaces, called cavities, that are separated by a scanty connective tissue stroma. More than 50% of all haemangiomas occur in the head and neck region with the tongue, buccal mucosa, lips and palate most commonly involved. The clinical appearance of these lesions is inconsistent; the size ranges from pinpoint to several centimetres in diameter, the colour ranges from bright red to purple and the position ranges from deep to superficial. The surface can appear flat or as a raised nodular mass. Superficial soft-tissue haemangiomas have a predilection for the head and neck while deep-seated haemangiomas appear more frequently in the trunk and lower extremities. Haemangiomas do not metastasise, but rather proliferate or involute with time.

The indications for therapy include acceleration of tumour growth, uncontrollable pain, gross functional impairment, local skin necrosis, thrombocytopenia, cosmetic deformity and suspicion of malignancy. The treatment options for haemangioma include surgical removal, injection of sclerosing agents, cryotherapy, radiotherapy and corticosteroids. The treatment depends on the location, size and age of the patient, sub-type of haemangioma, the presence or likelihood of complications, parental preference, and finally, expertise of the physician. In the present case series, different types of treatment modalities were given to both the cases diagnosed with haemangioma depending on the above-mentioned factors.

In case 1, a surgical approach was considered because there were well-circumscribed malformations of moderate size in which the possibilities of anatomic and functional restoration were maximal.
Among the different sites of head and neck haemangiomas, the tongue requires special consideration because of its susceptibility to minor trauma and consequent bleeding and ulceration, swallowing difficulties and breathing problems. If the lesion is accessible surgically, surgical excision is the gold standard treatment. However, surgical treatment of more extensive lesions can often lead to loss of motor function, nerve damage and massive bleeding.

In case 2, sclerotherapy was indicated as an alternative method of treatment for haemangioma. Different sclerotic agents have been used with varying degrees of success such as sodium tetradecyl sulphate, sodium morrhuate, sodium citrate, invert sugar, boiling water and sodium pylluate. In the present case 2 patient, 3% setrol (sodium tetradecyl sulphate) mixed with lignocaine HCL once a week for 3 weeks was injected intralesionally. It causes localised inflammatory reaction, obliteration of the haemangiomatous space, subsequent fibrosis of the endothelial spaces and regression of the lesion without affecting the bone. The advantages of the sclerosing agent included that it is simple and inexpensive; no loss of blood, no hospitalisation is required whereas the most common side effects are burning sensation, pain, swelling, redness and superficial ulcerations. Sclerotherapy can also be used to reduce the size of the lesion preoperatively as support to surgery or as a postoperative complement.

In both cases, no similar lesions were found in the other body systems whereas 20% of the haemangiomas were present at more than one site.

**Conclusion**

There is no ‘one-size-fits-all’ approach to the management of haemangiomas. The decision to treat depends on the individual patient and the specific clinical scenario at hand. Despite different recommended modalities in managing haemangiomas, the main goal of the treatment should be toward reducing the symptoms with minimal functional and aesthetic impairment as well as better patient compliance. The most adequate form of treatment depends on the case in question and each patient should be individually evaluated within the various contexts identifying the proper and most appropriate treatment modalities. However, when treatment is indicated for oral haemangiomas, appropriate care needs to be rendered to maintain oral functions as well as to concentrate on the aesthetic needs of the patient. Because there are no gold standard therapies and a paucity of randomised clinical trials in support of these interventions, the clinical experience of the oral physician is extremely helpful in guiding decisions about when, why and how to treat haemangiomas.

**Key points or key messages**

- As it is the most common tumor in infancy, health care physicians should ensure about spreading awareness and realizing goals with strategies.

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**Conflicts of interest**

There are no conflicts of interest.

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