

Hemangiolympangioma of buccal mucosa: Report of a rare case and review of literature on treatment aspect

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ABSTRACT

Hemangiomas are lesions that are not present at birth. They manifest within first month of life, exhibit a rapid proliferative phase, and slowly involute to non-existent. Hemangiomas of the oral cavity are not common pathological entities, but the head and neck are common sites. Furthermore some time histologically lymphatic channels may be evident in hemangiomas, and then it will be categorized according predominant component, e.g.; hemangiolympangiomas or lymphangiohemangiomas, vice versa is true for lymphangiomas. Most true hemangiomas involute with time, but 10-20% of true hemangioma in completely involute and required post- adolescent ablative treatment. In the present article we are reporting a case of hemangiolympangiomas of left buccal mucosa in an 18-year-old male patient. We have also reviewed various treatment modalities and their clinical implication.

Key words: Buccal mucosa, hemangiolympangiomas, hemangiomas, oral cavity

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INTRODUCTION

Although hemangioma is considered one of the most common soft tissue tumors of the head and neck,^[1] it is relatively rare in the oral cavity and uncommonly encountered by the clinicians. They may be cutaneous, involving skin, lips and deeper structures; mucosal, involving the lining of the oral cavity; intramuscular, involving masticator and perioral muscles; or intraosseous, involving mandible and/or maxilla.^[2-4]

Though previously termed “angiomas” or vascular “birthmarks”, vascular anomalies are divided into two main categories: Vascular tumors and vascular

malformations. Infantile hemangiomas comprise the majority of vascular anomalies and are considered the predominant vascular tumor type composed of rapidly proliferating endothelial cells.^[5] Blood vessel architecture is incomplete and surrounded by hyperplastic cells in hemangiomas and other vascular tumors. In contrast, vascular malformations do not contain hyperplastic cells, but consist of progressively enlarging aberrant and ectatic vessels composed of a particular vascular architecture such as veins, lymphatic vessels, venules, capillaries, arteries or mixed vessel type. The latter comprises lymphangiomas or lymphatic malformations which are congenital collections of ectatic lymph vessels that form endothelial lined cystic spaces.^[6]

Occasionally, channels may be filled with blood, a mixed hemangiolympangioma, an uncommon developmental anomaly with a propensity to invade underlying tissues and to recur locally, distinguishing it from the simple lymphangioma or hemangioma.^[7] Although histologically it is a benign disorder, local

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invasion into the muscle, bone, and underlying tissue can lead to severe deformity.^[6]

Management of hemangiomas and the treatment of choice depend on several factors including the age of the patient and the size and extent of the lesions, as well as their clinical characteristics.^[8]

CASE REPORT

An 18-year-old male patient, reported to Department of Oral and Maxillofacial Surgery, Trivandrum, Kerala, India, in May 2011, with the complaint of reddish swelling over left side of the upper lip with occasional bleeding while brushing since last 3-4 years. Patient noticed swelling over the left side of upper lip region 3-4 years back. Patient had repeated episodes of bleeding while brushing teeth or on mechanical trauma to the lesion. Patient visited private hospital, where he was kept on vitamin tablets and no other medications as history suggested by patient. The swelling was initially small papular which increased slowly to a size of 2×1 cm approximately in greatest dimension in last 2 years, after that there was no further change in swelling. At the time of reporting patient was having complaint of occasional bleeding from the lesion. Past medical and family history was non-contributory. Local examination revealed reddish multiple papular lesion over left side of buccal mucosa extending from 1cm behind the left corner of mouth in relation to 24, 25 of size approximately 2×1 cm [Figure 1]. There was no ulceration. On palpation the lesion was soft in consistency, non-tender, with smooth borders and was pulsatile in nature. There was no obvious increase in temperature of lesion. No obvious feeder vessel was identified. No obvious bleeding was identified on palpation of lesion. There was no associated lymphadenopathy. Provisional diagnosis of hemangioma left buccal mucosa was made. To rule out intracranial and bony involvement CT head and orthopantomogram (OPG) was taken, subsequently no abnormalities has been detected [Figures 2 and 3]. Excisional biopsy carried out under local anesthesia in proper aseptic conditions. Intraoperatively no obvious feeder vessel was identified. Mild to moderate bleeding was encountered and was managed by cauterization. Wide complete excision was done and surgical site was closed primarily. Gross specimen was soft in consistency and reddish in color and approximately of size 1.5×1 cm [Figure 4]. Specimen was sent for histopathological examination. Hematoxyline and eosine stain section shows hyperplastic stratified squamous epithelium which is extensively proliferating in to underlying moderately collagenous connective tissue stroma. There are numerous endothelial line

vascular spaces through the stroma and extending in to submucosa. Intermixed with large vascular channels dilated lymphatic vessels also seen. Endothelial cells proliferation and forming vascular channels were also evident. There is minimal inflammatory infiltrate [Figure 5]. Histopathologically diagnosis of hemangiolympangoama was made. Patient is on continuous follow-up no recurrence was noted till date.

DISCUSSION

Hemangiomas are a common soft tissue tumor that often congenital or develop in the neonatal period and grow rapidly. They usually cover a large site, may be macular or raised and usually resolve progressively in childhood.^[1,9] They may occur in the oral and maxillofacial region including gingiva, palatal mucosa, lips, jawbone, and salivary glands.^[3,10-14] The occurrence of hemangioma with its primary location on gingival tissues seems to be extremely rare. There are many clinical features of capillary hemangioma such as asymmetry of the face, spontaneous bleeding, pain, mobility of teeth, blanching of tissue, pulsation, expansion of bone, paresthesia, early exfoliation of primary teeth, delayed eruption, root resorption, and missing teeth.^[3,10,14] Hemangiomas are associated with various syndromes, Rendu-Osler-Weber syndrome (autosomal dominant inheritance, multiple telangiectasias, occasional GI tract involvement, occasional CNS involvement), Sturge-Weber-Dimitri syndrome (non-inherited and non-familial, portwine stain, leptomeningeal angiomas), von Hippel-Lindau syndrome (genetic transmission variable, hemangiomas of cerebellum or the retina, and cyst of the viscera).^[15] In present case intracranial and bony involvement is ruled out by CT Head and OPG.

Hemangiomas may mimic other lesions clinically, radiographically and histopathologically. The

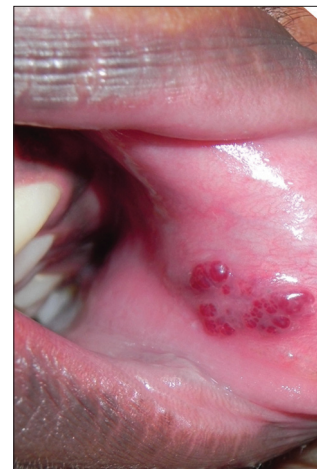


Figure 1: Clinically reddish multiple papular lesion left buccal mucosa

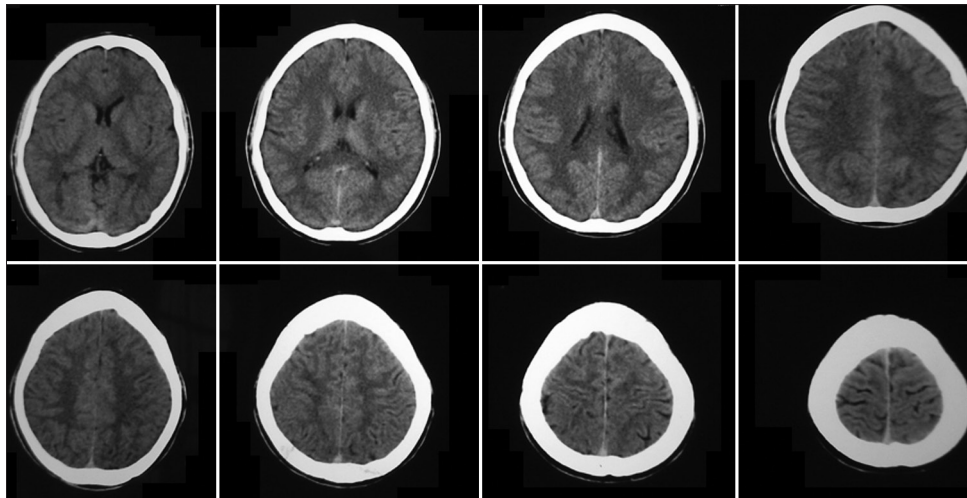


Figure 2: CT head with no detected abnormality



Figure 3: Normal OPG



Figure 4: Gross specimen

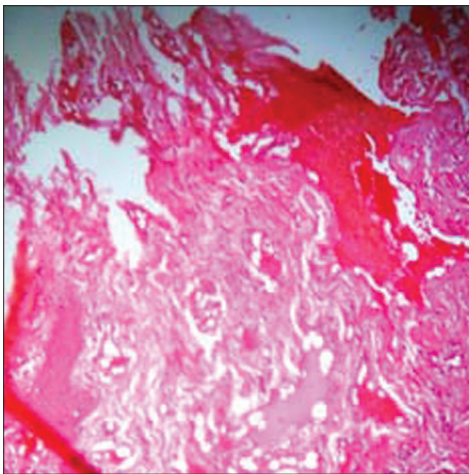


Figure 5: Hematoxyline and eosine stained section showing features of hemangiolympangioma

differential diagnosis of hemangiomas includes pyogenic granuloma, chronic inflammatory gingival hyperplasia (epulis), epulis granulomatosa, varicocell, talengetasia, and even with squamous cell carcinoma. The most common vascular proliferation of the oral mucosa is the pyogenic granuloma. This is a reactive lesion that develops rapidly, bleeds easily and is usually

associated with inflammation and ulceration. Clinically, it is often lobulated, pedunculated and red to purple and it may be hormone sensitive. Histopathologically, hemangioma exhibits a progression from a densely cellular proliferation of endothelial cells in the early stages to a lobular mass of well-formed capillaries in the mature phase, often resembling the pyogenic granuloma without the inflammatory features. Some cases intermingled lymphatic channels are also evident. Diagnosis is given according to predominant component, e.g.; hemangiolympangioma and/or lymphangiohemangioma, this particular terminology is also true in case of lymphangiomata.^[6] As in our case there were intermingled lymphatic channels, but predominant component was of hemangioma, so diagnosis of hemangiolympangioma was made.

Precise diagnosis of the type of vascular lesion is important because it may influence treatment considerably. Angiographic studies are not strictly demonstrated for diagnosis of hemangiomas, and are

utilized only to define the size and the extent of the lesion.^[10,14] These are more complicated procedures than histopathological evaluation, have a higher morbidity, and may cause undesirable side-effects. CT and MRI of these lesions have more recently been demonstrated, and have been successfully utilized for the diagnosis of hemangiomas, as for other lesions of soft tissues.^[16,17] In the case presented here, treatment of the hemangiolympangioma was done by wide surgical excision and primary closure. In present case no intraoperative complication encountered. The healing after primary closure was uneventful [Figure 6].

Various treatment modalities has been advocated, e.g.; Surgical excision, for those lesions not amenable to surgery other therapy such as intralesional injection of fibrosing agents, interferon a-2b, radiation, electrocoagulation, cryosurgery, and laser therapy, embolization may be used.^[10,18,19] Attempts to remove hemangiomas using surgical excision may lead to serious medical problems such as heavy bleeding. In addition, postoperative recurrence may encounter.^[2,10] In presented case no such complication encountered.

Bogdan *et al.*^[20] aimed a study to evaluate the efficacy of the 980 nm diode laser and Er:YAG laser for photocoagulation treatment of oral and maxillofacial hemangioma and vascular malformations. Seventy consecutive patients with either hemangioma or low-flow vascular malformations of the head and neck treated with laser photocoagulation were comprised in the study. Long-term follow-up demonstrated regression of the lesion in all patients with good aesthetic results. The range of reduction in size varied between 45% and 95% and no complications or reperfusion of the lesions was noted. Photocoagulation with diode laser or Er:YAG laser of hemangioma and vascular malformations is an effective treatment for correctly selected patients. They concluded that properly applied, these techniques can achieve reduction in size of these lesions without compromising function and cosmetics.

Bonet-Coloma *et al.*^[21] conducted an observational retrospective study, reviewing medical records with clinical diagnosis of haemangioma between 1990 and 2006 at the Children's Maxillofacial Surgery Service of the Hospital Universitario la Fe, Valencia. The study included 28 patients (19 females and 9 males) with a mean age of 4.27 years (range 0-14 years). Nine were congenital haemangioma. The most frequent location of oral haemangioma was in the lip with 23 cases, followed by three cases in the tongue and 2 in the buccal mucosa. The mean diameter of the lesion was 1.67 cm (range 1-3 cm). The mean duration of the lesion was 6.3 months (range 1 month to 5 years). Of the 28 haemangiomas, 13 were surgically removed, 2 were treated with

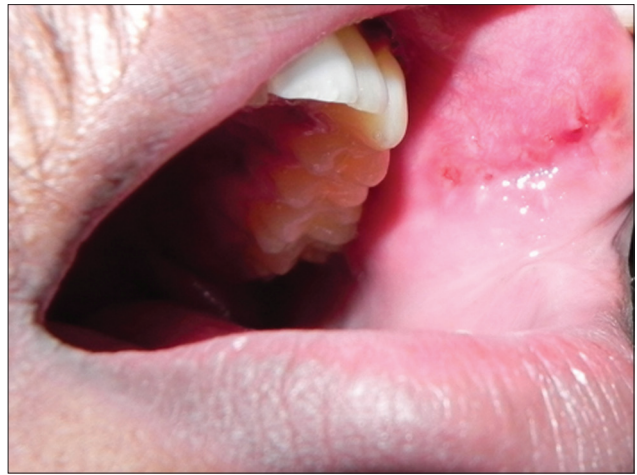


Figure 6: Post-operative uneventful healing

embolization and 13 disappeared spontaneously. The mean follow-up was 2.7 months (1-8 months). There were no cases of recurrence. Haemangiomas usually present in children, and can be seen from birth. They have a predilection for females. They are uncommon in the oral cavity. In the oral region, the most common location is the lip. Most congenital haemangioma regress spontaneously without treatment. The treatment of choice was the surgical excision in their study.

Plasma knife (PK) is a new treatment modality for excision of these lesions. PK technology provides an electrically conducting site by means of obtaining the use of controlled radiofrequency energy, and by using the intra and extracellular fluid. The active zone on the device provides an ionized layer by means of radiofrequency waves. Dense kinetic energy divides, demolishes and evaporates the constructional element tissue. During this procedure, there is very little thermal damage to the surrounding tissue. Because it requires no additional saline supply for the operation according to the thermal procedure, its tip temperature remains a cool 70-80°C. Therefore, it provides a pink viable resection bed. PK is developed for tonsillectomy, adenoidectomy, uvulopalatopharyngoplasty, thyroidectomy and excisions of the head and neck masses.^[22,23] They used a 90% coagulation and 10% cut mode choice for the PK excision of the haemangioma of the tongue. It was shown that the PK was capable to control the bleeding. This method is consequently a safe and suitable surgical choice to be applied for superficial and small sized haemangiomas of the tongue. In their case, there was no swelling of the tongue, bleeding nor pain postoperatively. There was also no problem of speaking and swallowing. The experience that we gained from this case is that the PK surgery would be a suitable treatment modality for the localized and superficial haemangiomas of the tongue. PK can also be combined with other treatment modalities in larger lesions.^[24]

In conclusion hemangiomas are not common in oral cavity. All suspected cases should be properly evaluated to rule out intracranial, bony and systemic involvements. Yet surgical excision is the mainstay of the treatment. Patient should be followed postoperatively to rule out complication and further long-term follow-up to rule out recurrence. As in present case no recurrence has been noted after approximately one year of follow-up.

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