



Case Report

Cavernous Hemangioma of Buccal Mucosa: A Rare Case Report with a Reappraisal of Differential Diagnosis & Review of Literature

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ABSTRACT

Hemangiomas are common tumors which are qualified as benign lesions as there is proliferation of dilated blood vessels. These are present at birth & gradually increased in size & by puberty involutes but few lesions persist. These lesions are clinically & histopathologically differentiated from other soft tissue lesions as the treatment modes are different for hemangiomas & other soft tissue lesions. For the dentist these have considerable importance as sometimes if there are intraosseous & if extraction is attempted can lead to severe bleeding. So knowledge of these is important for appropriate clinical management.

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Introduction

The category of vascular anomalies encompasses heterogeneous groups of lesions¹. In this class mainly two entities are important that is Hemangiomas & vascular malformations^{1,2}. The former–Hemangiomas are innocuous vascular lesions comprising of heterogeneous group of lesions that share similar histological features¹. The terminology hemangioma was first coined by Szejder *et al* in 1973 & was called “Haemorrhagic Hemangioma². Hemangiomas are benign proliferation of dilated blood vessels & capillaries¹⁻³. Hemangiomas are the most common soft tissue tumors of head & neck in children, increased in size till puberty or adulthood & then regress³⁻⁶. But few lesions can persist & are seen in older individuals^{2,7,8}. On few occasion these lesions are congenital & persist throughout life⁷. These lesions are commonly located on skin, lips, deeper tissues (intramucosal & intramuscular) & in bones (maxilla or mandible)^{1,4,9-11}. Females are predominantly affected than males¹⁻⁴. Mullikan & Glovack in 1982 proposed a classification to classify the vasoformative errors into hemangiomas & vascular malformation¹⁻³. Hemangiomas are further classified based on size of vascular spaces into four types that is capillary, cavernous, mixed & sclerosing¹⁻⁴. Based on vessel type it is classified into arterial (low flow) & arteriovenous (high flow)³. Clinically hemangiomas are soft, sessile or pedunculated painless growth with deep red to dark blue color with size ranging from small to larger lesions, the surface of which is smooth or lobulated^{1-4,7,8}. On palpation, these lesions exhibit blanching without bruits or pulsations. Intraorally these are commonly found on gingiva⁴, palate¹ & tongue³.

Here we present a case of Hemangiomas on buccal mucosa which is a rare site for developmental of Hemangiomas.

Case Report

A 26 year male patient reported to the department of oral medicine with a chief complaint of swelling on the right buccal mucosa since 3-4 years. Patient gives history of the swelling which he observed first 10 years back. At that time it was small in size & gradually increased in size to attain the present size. There is no pain or discomfort associated with the lesion.

The general examination of patient was normal for his age with no defect in gait or height. The patient medical & dental history was non contributory.

Intraoral examination revealed a single large sessile, smooth irregular painless swelling measuring about 3x4 cms, dark blue in color present on right side of buccal mucosa [Figure 1]. There is no pain, bleeding associated with the lesion. The borders of the lesions were well defined. On palpation the lesions was soft & blanch on application of pressure. The differential diagnosis of hemangioma or vascular malformation was pinned down to hemangioma when the auscultation exhibited no thrill or bruit.

Correlating the history & clinical findings a provisional diagnosis of Hemangiomas was arrived at.

The treatment protocol for the management included administration of sclerosing agent into the lesion & was kept under watch for period of 1 week. No change was observed in size & appearance was noted. Under local anesthesia surgical excised was carried out & the sutures were placed. During the procedure, bleeding was noted. The healing was uneventful.

The specimen was submitted for histopathological examination. The microscopic examination revealed a large sinusoidal spaces lined by flat endothelial cells engorged with red blood cells were noted [Figure 2].

Discussion

In the category of the soft tissue tumors Hemangiomas are common lesions that are developmental or congenital & grow rapidly in size till puberty & then regresses¹⁻³. Hemangiomas are normally seen in childhood⁷, adults³ but few lesions persist later in life^{3,5}. These lesions adapt an indolent course⁴.

The exact etiology is not known but few authors have hypothesized that angiogenesis plays a vital role in vascular excess present¹. Cytokines like basic fibroblast growth factor (bFGF) & vascular endothelial growth factor (VEGF) are acknowledging inducing the process of angiogenesis^{1,2}. So in this pathogenesis of this vascular lesion there is an imbalance in the factors that stimulates angiogenesis & the factors which conquer angiogenesis (Gamma interferon, tumor necrosis factor-beta, transforming factor-beta)¹.

With respect to the gender, observation made by Alparslan Dilsiz *et al*², Krishna Kripal *et al*³, Nadeem Jeddy *et al*⁴ shows that females are more commonly affected than males, but in our case the patient was male.

Kocer U *et al*⁹, Acikgoz A¹⁰ suggested that head & neck region is common site that is affected. Comparatively oral site is less commonly involved but if affected gingiva followed by lip, tongue & palate are chiefly involved. Our case is contrary to the observation made by above authors & in our case buccal mucosa was involved.

Alparslan Dilsiz *et al*², Krishna Kripal *et al*³, Nadeem Jeddy *et al*⁴ in their study on hemangiomas noted that clinically these lesions were soft, sessile or pedunculated, smooth or lobulated, painless, purple or deep dark blue in color with size varying from small size to a larger lesion. Our observations are similar to the findings noted by above authors. On palpation, these

lesions blanch & don't show bruits or thrill^{8,11}.

Histologically we see mostly capillary, cavernous, mixed & sclerosing type⁵⁻⁷. Here in our case we noted cavernous type of hemangiomas with large sinusoidal spaces lined by flat endothelial cells that were engorged with red blood cells.

Differential diagnosis

Clinically & histopathologically hemangiomas can mimic many other soft tissue tumors like the vascular malformations, pyogenic granuloma, epulis, varicosities, OSCC, Kaposi sarcomas²⁻⁴. The differential diagnosis has a vital role to play in these lesions as it can dictate the treatment option & the outcome of the disease^{4,5}.

Pyogenic granuloma is a reactive lesion which appears suddenly^{1,3}. The etiology for this can be calculus, broken teeth, trauma, foreign body & hormonal changes⁴. One important point to be remembered in case of pyogenic granuloma is the age of occurrence which is seen mostly in 2nd & 3rd decade of life^{1,3,4,12,13} where as the hemangiomas occur in childhood & involutes by puberty.

Vascular malformations are present from birth mostly affecting both males & females in equal frequency¹². The clinical behavior is quite different from the hemangiomas as these vascular malformations continuous inc in size throughout life & the basic fault in these lesions is that there is structural defect in the blood vessel where as in the hemangioma there is proliferation of delayed blood vessels^{1,2,12,13}.

Varicosities are the benign proliferation of blood vessels usually seen in old age & once formed don't regress in life^{12,13}.

Epulis are the inflammatory gingival hyperplasia which is seen mostly in 2nd -4th

decade of life with specific etiology related to denture injuries or ill fitting dentures¹². Oral squamous cell carcinoma on other hand the etiological factor as tobacco & the growth is seen in 3-5 decade of life^{1,3,12}.

Kaposi sarcoma is an unusual vascular neoplasm & current evidences suggest that Human Papilloma Virus is the main causative factor^{12,13}. It manifest clinically in four different forms- classic showing the bluish macules & papules which gradually develop into nodule^{12,13}. The endemic type (African) similar to the classic variety predominantly seen in African children & the iatrogenic-immunocompromised associated seen in organ transplants patient. The fourth variety seen in AIDS patients^{12,13}.

Histologically pyogenic granuloma have lobular arrangement of capillaries with specific inflammatory component of neutrophils, lymphocytes & plasma cells where a sin hemangiomas mostly the inflammatory infiltrate is absent unless it is infected or ulcerated^{4,5,12}.

Vascular deformations have vascular defects which maybe either capillary, venous, arterial or arteriovenous type with defect in blood vessels^{1,12}.

In Varicosities mostly veins are involved which become swollen & tortuous^{12,13}.

Epulis shows a hyperkeratotic hyperplastic tissue with arcading reteridges with inflammatory infiltrate^{1,4,5,12,13}.

Oral squamous cell carcinomas has dysplastic features plus the keratin pearl formations which can be categorized as well- differentiated, moderate differentiated & poorly differentiated¹².

Kaposi sarcoma depending upon the stage shows the corresponding histological features i.e. the macular stage shows the bland endothelial cells with engorged blood vessels, plaque stage-proper blood vessels

with spindle cells component & the nodule shows more of spindle cell component^{12,13}.

There few syndrome that are associated with these lesion like Osler-Weber- Struge syndrome & Struge – Weber syndrome with prominent hemangiomas, ocular & cerebral hemangiomas^{3,4}. These lesions don't undergo spontaneous involution as hemangiomas do⁴. These are further classifies as flat, telengiectasia, stellar & senile variant¹⁻⁴. Precise diagnosis of the type of vascular abnormality is paramount as it decides the treatment & its outcome. Angiographic studies are not strictly demonstrated for diagnosis of hemangiomas¹⁴. For the diagnosis of hemangiomas, recent advance techniques like MRI & CT scan have shown good results^{15,16}. The treatment of hemangiomas varies depending upon the clinical manifestation & the anatomic location. Surgical excision is the treatment of choice for many¹⁷. For those lesions not amenable to surgery, other therapy such as intralesional injections of fibrosing agents, interferons, cryosurgery, laser therapy, electro coagulation & radiation may be utilized¹⁸.

Conclusion

The identification of hemangioma is challenge as these lesions clinically resembles other entities like the vascular malformation, pyogenic granuloma, epulis, varicosities & oral squamous cell carcinoma. Extensive work up has to be done to arrive at the accurate diagnosis taking into account the differential diagnosis & the latest technology in diagnosis of the lesions. For clinical practitioner who has limited knowledge about these lesions diagnosis is a uphill task & it involves lot of risk during the treatments like the extractions & periodontal surgery as it results in profuse bleeding creating an emergency situation. In depth analysis of clinical growth pattern &

histological type has to be kept in mind to yield targeted therapeutic treatment & abbreviate social embarrassment to the individual.

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Figure 1. Intraoral examination reveals single large sessile, smooth irregular painless swelling of Right Buccal Mucosa

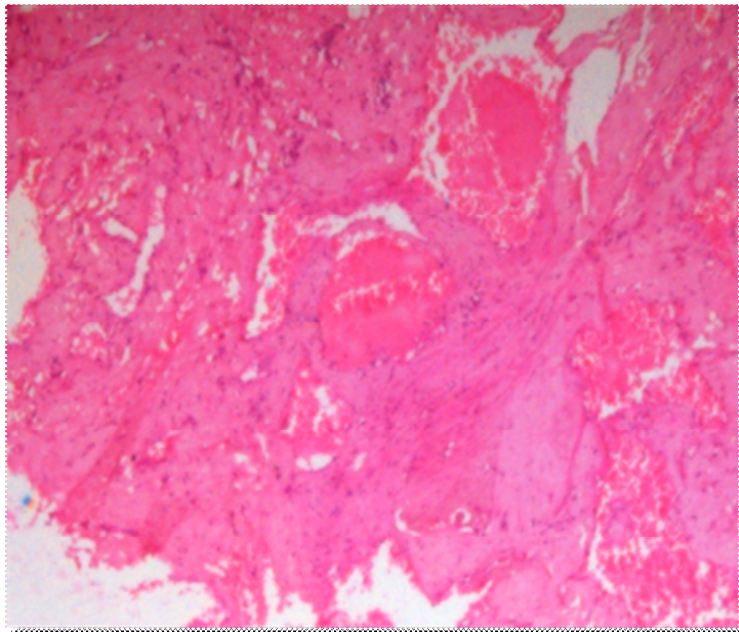


Figure 2. Photomicrograph exhibiting large sinusoidal spaces lined by flat endothelial cells engorged with red blood cells