



Capillary and Cavernous Hemangiomas of the Maxilla: Case Report of Two Patients and A Review of the Literature

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Abstract

Hemangiomas are the most common benign vascular lesions of the head and neck region. However, their occurrence in the oral cavity is relatively rare, especially in oral soft tissue. Hemangiomas are endothelial cell proliferations, followed by gradual involution tumors with unique biologic characteristics that they grow rapidly regress slowly and have very less recurrence. This case report present a case of central hemangioma in a female patient in the maxillary canine-premolar region of unusual size, which is uncommon and a case of cavernous hemangioma in buccal and palatal area of male patient. Even though the lesion has benign growth yet,detailed description, clinical and histopathological features along with new treatment modalities are hugely important for a dental profession.

Introduction

The term hemangioma has traditionally been used to describe a variety of developmental vascular anomalies. Hemangiomas are benign, enlarged, vascular hamartomas that may be seen in any soft tissue or bony intra-oral location. They occur early in life and somewhat more commonly in women than in men [1,2]. Soft tissue hemangiomas occur commonly in the dorsum of the tongue, gingiva and buccal mucosa. Vascular lesions of the face are not very common. These lesions are a perplexing group of problems that over the years have generated a significant debate and confusion as regards their terminology and classification. Descriptive anatomic, pathologic and embryologic classification scheme have been devised however generally have not offered the clinician significant guidance for treatment. The classification developed by Mulliken and Glowacki in 1982 is based on the cellular kinetics of anomalous vessels, providing a diagnostic and therapeutic approach based on the biologic behaviour of the lesion. In this classification, two entities exist: 1) hemangiomas and 2) vascular malformations [1,2]. The International Society for the Study of Vascular Anomalies (ISSVA), in 1996, approved a classification system modified from the one proposed by Mulliken, Glowacki (1982) [3]. The diseases were subdivided into (a) tumors: hemangioma (HEM), pyogenic granuloma, rapidly involuting congenital hemangioma, noninvoluting congenital hemangioma, hemangiopericytoma, tufted angioma and kaposiform hemangioendothelioma; and (b) vascular malformation (VM) [4].

Wassef “et al.” [5] updated official ISSVA classification of vascular anomalies in 2015. The General biological scheme of the classification is retained. The section on tumors has been expanded and lists the main recognized vascular tumors, classified as Benign, locally aggressive or borderline, and malignant. A list of well-defined diseases has been included under each generic heading in the “Simple Vascular Malformations” section. Two new sections were created: one dealing with the malformations of individually named vessels (previously referred to as “truncular” malformations); the second groups lesions of uncertain or debated nature (tumor versus malformation) [5]. Hemangiomas are considered to be benign tumors and has a predilection towards of infancy that are characterized by a rapid growth phase with endothelial cell. It presents as a red macula, papule or nodule, depending on the congestion degree and on how deep it is in the tissue. Although HEM is a benign lesion, in some cases, it may lead to compression of surrounding structures, formation of fissures, ulcers or hemorrhages, and functional and aesthetic problems [6,7]. Oral HEM can be found in the lips, tongue or buccal mucosa. It is more common in white female, in twins and in premature

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Figure 1: Erythematous growth extending to palatal region.



Figure 2: Intra oral picture showing well defined erythematous growth.

infants. On the other hand, vascular malformations are structural anomalies of blood vessels without endothelial proliferation [1-3]. Clinically, they are similar to HEM; however, they are always present at birth and grow as the patient physically develops [3]. It does not spontaneously regress, remaining stable throughout life. Frequently, bone involvement is present as a radiolucent, multilocular and well-circumscribed image [6]. The few studies on benign oral vascular lesions frequently do not distinguish between oral HEM and VM, or regard oral VM as a histological type of HEM, known as arteriovenous hemangiomas [8].

Vascular Malformations may be single vessel forms (capillary, arterial, lymphatic, or venous) or a combination. They are designated according to the predominant channel type as capillary malformations, lymphatic malformations, venous malformations, arteriovenous malformations, and complex forms such as capillary lymphatico-capillary lymphatico-venous malformation. Malformations with an arterial component are rheologically fast flow, while the remainder are slow-flow. Hemangiomas are classified on the basis of their histological appearance as capillary, cavernous or mixed. Capillary hemangiomas are composed of many small capillaries lined with a single layer of endothelial cells supported in connective tissue stroma of varying density. Cavernous hemangiomas are formed by large, thin-walled vessels or sinusoids that are lined with a single layer of endothelium and are separated by thin septa of connective tissue. Mixed hemangiomas consist of both capillary and cavernous components [8].

Case Presentation

Case 1

A female patient of 35 years age reported to the Department of



Figure 3: Orthopantomogram showing ill defined radiolucency in periapical region of 23-25 region.



Figure 4: Histopathology of patient.

Oral medicine and radiology of Government Dental College and hospital Jammu with the chief complaint of swelling in the upper gingival area for the past 2 months which was slowly progressive nature. There were no relevant contributory history to our case report. Patient was a Housewife, Vegetarian and had no history of any tobacco usage of any form. On clinical examination no significant extra-oral finding and no visible facial asymmetry was noted. Lymph nodes were non palpable. Intra-orally a well defined erythematous growth with distinct borders, arising from the gingival aspect of the Maxillary Second Quadrant's Central incisor to second pre molar region was seen. The same lesion of 2.5 cm x 1.5 cm in size extended to palatal aspect of the maxillary Premolar area (Figure 2). On palpation the growth was firm in consistency with blanching and bleeding on provocation was seen. Non tender, no ulceration and no pulsation or bruit was noted (Figure 1 and 2).

Radiographical investigation: Intra-oral periapical (IOPA) X-ray revealed the presence of an ill-defined radiolucency involving the periapices of 23–25 with slight displacement of regional teeth. There was no significant bone destruction (Figure 3). Based on the clinical and radiological findings, a provisional diagnosis of pyogenic granuloma was made. The differential diagnosis for this intra oral lesion was peripheral ossifying fibroma and peripheral giant cell granuloma. Incisional biopsy of both the soft tissue mass and underlying bone was performed under local anesthesia. The tissue was preserved in 10% formalin and sent for histopathological examination.

Histopathologically: The excised specimen showed few large endothelial cells lined blood vessels and numerous proliferating endothelial cell lined capillaries. Containing RBC's in a lobular pattern segregated by fibrous septa in the connective tissue stroma. Chronic inflammatory infiltrate composed predominantly of Plasma cells and Lymphocytes and few extravasated RBC's were also present. Thus, this was suggestive of capillary hemangioma (Figure 4).



Figure 5: Multilobulated swelling present over the right buccal.



Figure 6: Vascular formations extending to palatal region.



Figure 7: Lateral cephalogram of patient.

Case 2

A male patient of 45 years age reported to the Department of Oral Medicine and Radiology of Government Dental College and Hospital Jammu with the chief complaint of swelling present over the right side of his mouth since 2 months. There was no significant medical, dental and family history given by the patient. The patient was a businessman by profession, non vegetarian and had no history of any tobacco usage of any form. Clinical examination of patient reveals no extra oral swelling, no facial asymmetry and no palpable lymph nodes. Intra oral examination revealed Multilobulated swelling present over the right buccal mucosa area (Figure 5). There is irregularity in shape, extending from retro-commercial area upto retromolar area, Around 4 cm x 3 cm, 2 cm x 3 cm & 2 cm x 2 cm in size respectively. Overlying mucosa was reddish blue in color. (mention picture reference). A growth was seen on buccal mucosa with burrowing on the buccal vestibule Irregular in shape , overlying mucosa is deep red in color.



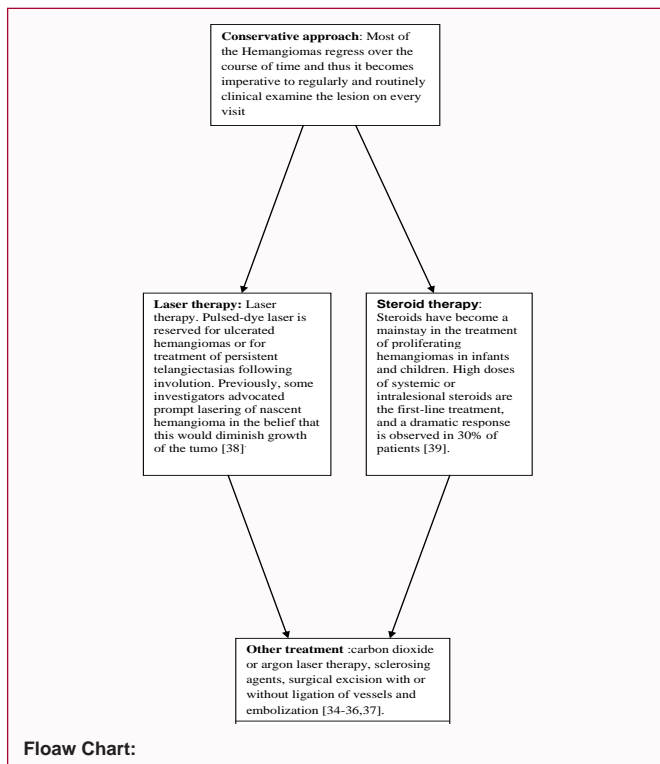
Figure 8: Histopathology of patient.

On palpation: Soft non - tender, easily bleeds on provocation. A flat macular lesion is present near the palatal region on the right side, blue in color around 2 cm x2 cm in size diffuse in shape (Figure 6). Radiographs were advised to rule out any bony lesion suggestive of central variety of hemangioma, or any malignancy or to identify a foreign body that should need to be removed with the lesion, however in our case the orthopantomogram as well as the lateral cephalogram. Didn't reveal any significant changes (Figure 7).

Histopathologically: The excised specimen showed stratified squamous keratinized epithelium with the connective. Tissue stroma showing large dilation of endothelial lined vascular spaces. Along with engorged Red blood cells. Deeper connective tissue layers showed chronic inflammatory cell infiltrates along with adipose and muscle cells, all this suggested of vascular malformations (Figure 8).

Discussion

The history of hemangioma dates back to 1843 when Liston first described the case of hemangiomas [9]. Later on cases of vertebral hemangioma (Virchow, 1867) [9] cutaneous hemangiomas (Kasabach and Merrit, 1940) were documented [10]. In 1973 Sznajder "et al." [11] described hemangiomas under the term "Hemorrhagichemangioma". However, there have been many conflicting reports over the nomenclature. Bormann first used the term hemangioendothelioma in 1899 [12]. In 1908, Mallory used the term malignant vascular tumor for this lesion [13]. Stout described the histological features of hemangiomas in 1944 [14]. Hemangiomas are the most common soft tissue tumor of childhood [15–17] and head and neck region is the most common site for hemangiomas development (about 60% of cases) [17]. Hemangiomas are the most common benign tumours of the head and neck in children, but their occurrence on the palatal mucosa is extremely rare [18]. In 80% of cases, hemangiomas occur as single lesions [19]. Moreover, capillary hemangiomas have a 3:1 female-male ratio and occur more frequently among Caucasians than other racial groups [19,20]. Also it has been documented that Hemangiomas are the most common tumors of infancy, occurring in 5% to 10% of 1-year-old children. They are much more common in females than males. The most common location is the head and neck region, which accounts for 60% of all cases. Eighty percent of hemangiomas occur as single lesions, but 20% of affected patients will have multiple tumors. Hemangiomas of the oral mucosa may be flat or raised, often multinodular, and distinctly reddish, blue, or purple [20,21]. Most oral hemangiomas are located on the tongue, where they are multinodular and bluish red. The multinodularity is racemose and diffuse. Tongue angiomas frequently extend deeply between the intrinsic muscles of the tongue. The lip mucosa is another common site for hemangiomas in children; these tumors are usually localized, blue, and raised. The aforementioned port-wine stain involves the facial skin and is flat and



magenta in color. Histopathologically, hemangiomas are classified as cavernous or capillary type according to their

vascular network. Early hemangioma is characterized by the presence of plump endothelial cells and indistinct vascular lumen and is often known as turbine or cellular hemangiomas [23]. Thickened multi laminated endothelial basement membrane with the ready incorporation of tritiated. Thymidine in endothelial cells and the presence of a large number of mast cells are noted in the Proliferating phase of hemangiomas [24]. As it matures, the endothelial cells become flattened and tiny capillary sized vascular spaces become prominent [24]. When the lesion involutes, the vascular. As spaces become more dilated (cavernous) and widely spaced [24]. There is little or no incorporation of tritiated thymidine in endothelial cells and normal count of mast cells are noted in the involuting phase [24]. Capillary hemangiomas are usually present at birth, while most cavernous hemangiomas occur in adulthood. Cavernous hemangiomas are less common than capillary hemangiomas in all other areas of the body except in the oral cavity. Almost all intraosseous hemangiomas of the facial skeleton are to be cavernous type [24].

The diagnosis of hemangioma is based on clinical history and physical examination. Imaging studies may be necessary to clarify and confirm the diagnosis, and in order to analyze the extent of the lesions by permitting an evaluation of their non-visible component as well as the affection of neighboring structures. The imaging techniques employed for hemangiomas include MRI, CT, CT with contrast media, ultrasonography and angiographic techniques (arteriography, phlebography) [25-32].

Syndromes associated with cavernous hemangiomas are Sturge Weber syndrome, Kasabach Merritt syndrome, PHACE (posterior fossa brain malformations, haemangioma of the face, arterial cerebrovascular anomalies, cardiovascular anomalies, eye anomalies, and sterna defects or supraumbilical raphe) syndrome [33].

The management of hemangiomas of the oral mucosa varies according to the age of the patient, the size of the lesion, the site of involvement and the clinical nature of the hemangioma. The range of treatment includes steroid therapy, carbon dioxide or argon laser therapy, sclerosing agents, surgical excision with or without ligation of vessels and embolization [34-36,37]. Larger lesions that extend into muscles are more difficult to eradicate surgically, and sclerosing agents such as 1% sodium tetradecyl sulfate may be administered by intralesional injection. These agents result in postoperative pain, and the patient must be managed with a moderate-level analgesic such as oxycodone or aspirin with codeine. Cutaneous port-wine stains can be treated by subcutaneous tattooing or by argon laser. However in our case 1, after all lab investigations and histo pathological reports were done, surgical excision of the lesion was performed under local anesthesia under all aseptic conditions. Because the lesion was localized and had no underlying pathology. There is been no recurrence of the lesion at 1-month, 6-month, or 1-year follow-ups as patient has been reviewed periodically. In our 2nd case intralesional triamcinolone acetonide (4 mg/mL) was started weekly upto 6 months. Patient had a definite response and showed great improvement. The patient has been on review (Flow Chart).

Conclusion

Haemangiomas are common benign vascular growth, but their occurrence in the oral cavity has been rare, thus it becomes imperative for dental professional to evaluate them clinically, histopathologically and undertake all necessary investigation. Dental professionals should be well versed with all the clinical and treatment modalities that are associated with hemangiomas and all necessary precautions should be taken before attempting surgical excision as the tissues may bleed profusely and unexpectedly.

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