Vascular neoplasms in the differential diagnosis of gingival growths

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ABSTRACT:

Neoplastic gingival enlargements include a variety of benign and malignant tumors, however, vascular tumors such as hemangiomas and lymphangiomas occurring primarily on the gingiva is an uncommon event. Hemangiomas are benign tumors of blood vessels which are classified into cavernous and capillary type based on histology. Similarly, lymphangiomas are benign, hamartomatous tumors of lymphatic vessels which do not communicate with the rest of the lymphatic systems. Both tumors show a marked predilection for head and neck region (hemangioma-60%; lymphangioma-50-75%); yet intra-oral occurrence especially on the gingiva is very rare. Thus, the aim of this paper is to present two cases, one each of hemangioma and lymphangioma of gingiva. It is also warranted that these tumors should be included in the differential diagnosis of gingival enlargement.

Keywords: *Gingiva; enlargements; hemangioma; lymphangioma; vascular*

INTRODUCTION

Benign vascular neoplasms are very common and most frequently occur in the skin. [1] This spectrum of tumors includes hemangioma, lymphangioma, angiomatosis, and hemangioendothelioma. At all sites, it is oftentimes difficult to determine whether benign vascular lesions are malformations, true neoplasms or, in some cases, reactive processes. Similarly, it remains essentially impossible to reliably distinguish blood vessel endothelium from lymphatic endothelium, which probably reflects the close functional and embryogenetic relationship between these cell types.[1]

Both hemangiomas and lymphangiomas show a marked predilection for head and neck region (hemangioma-60%; lymphangioma-50-75%).[2] Howbeit, occurrence of vascular with its primary location on gingiva is infrequent.[3] On the basis of the size of vascular spaces and histology, hemangiomas can be classified into capillary and cavernous types. [4] As with hemangiomas, it is ordinarily arduous to state whether lymphangiomas are true neoplasms, hamartomas, or lymphangiectasias. In actuality, this distinction is of little practical value because they are all benign lesions, and therapy is largely dictated by their location and clinical extent.[5] Most regard lymphangiomas as malformations that arise from sequestrations of lymphatic tissue that fail to communicate normally with the lymphatic system. Histological classification of lymphangiomas into capillary, cavernous or cystic subtypes is not used anymore. Alternatively, the all-inclusive term "lymphangioma" is preferred since the histological distinction is of little practical importance. [5]

Gingival enlargements or gingival over growths are the current accepted terms for increase in the size of the gingiva.[3] Neoplastic gingival enlargements include a variety of benign and malignant tumors, however, vascular tumors occurring primarily on the gingiva are rare. Clinically these tumors may closely resemble pyogenic granuloma, thus most of the time clinicians do not submit the excised tissue for histopathological examination. This may be one of the reasons of rareness of such tumors on gingiva. We hereby present two cases, one each of hemangioma and lymphangioma of gingiva and warrant that these tumors should be included in the differential diagnosis of gingival enlargement.

CASE HISTORY

CASE 1

A thirteen years old female patient presented with a chief complaint of a growth in the lower right back tooth region since two months. There were no associated symptoms such as pain or pus discharge. The lesion bled while brushing since last two weeks. Intra oral examination showed an ovoid swelling of about 2x2 cm extending from distal aspect of 44 to mesial aspect of 46 impinging upon the right mucobuccal space (Fig 1A). Surface was smooth and mucosa over the swelling was erythematous. On palpation, it was soft in consistency with well defined borders however; the superior aspect appeared more fibrous and showed indentations of the maxillary posterior teeth. It was compressible and blanched on palpation. A provisional diagnosis of pyogenic

granuloma was made. The phase I periodontal therapy was instituted including scaling and root planing with other oral hygiene measures. The patient was under a routine follow up of three weeks. When the lesion did not regress after the primary measures (Fig. 1B) it was decided to excise the lesion in toto under local anesthesia. The excised specimen was immediately fixed in 10% buffered formalin and sent for histological examination. Histologically, the submitted section showed proliferating endothelial cell in a lobular architecture (Fig. 1D-E). Some areas showed canalizing endothelial channels (Fig. 1E-F). The intervening stroma was fibro-cellular and tissue was covered by stratified squamous epithelium. Based on this a diagnosis of cellular hemangioma was given and it was further confirmed by CD34 positivity by immunohistochemistry (Fig. 1E, inset). Post operative healing was uneventful. There was no sign of recurrence after a follow up of 2 months.

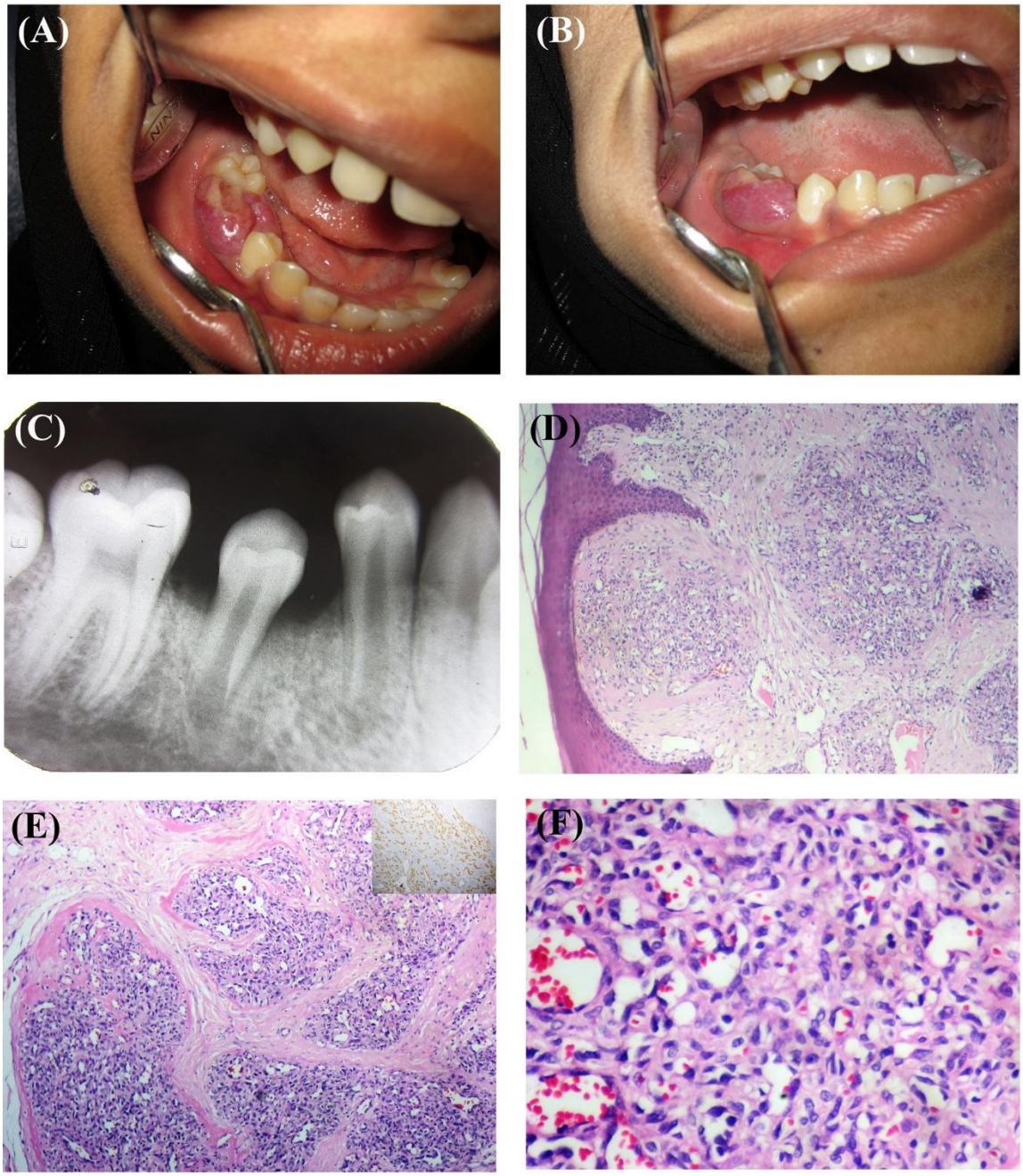
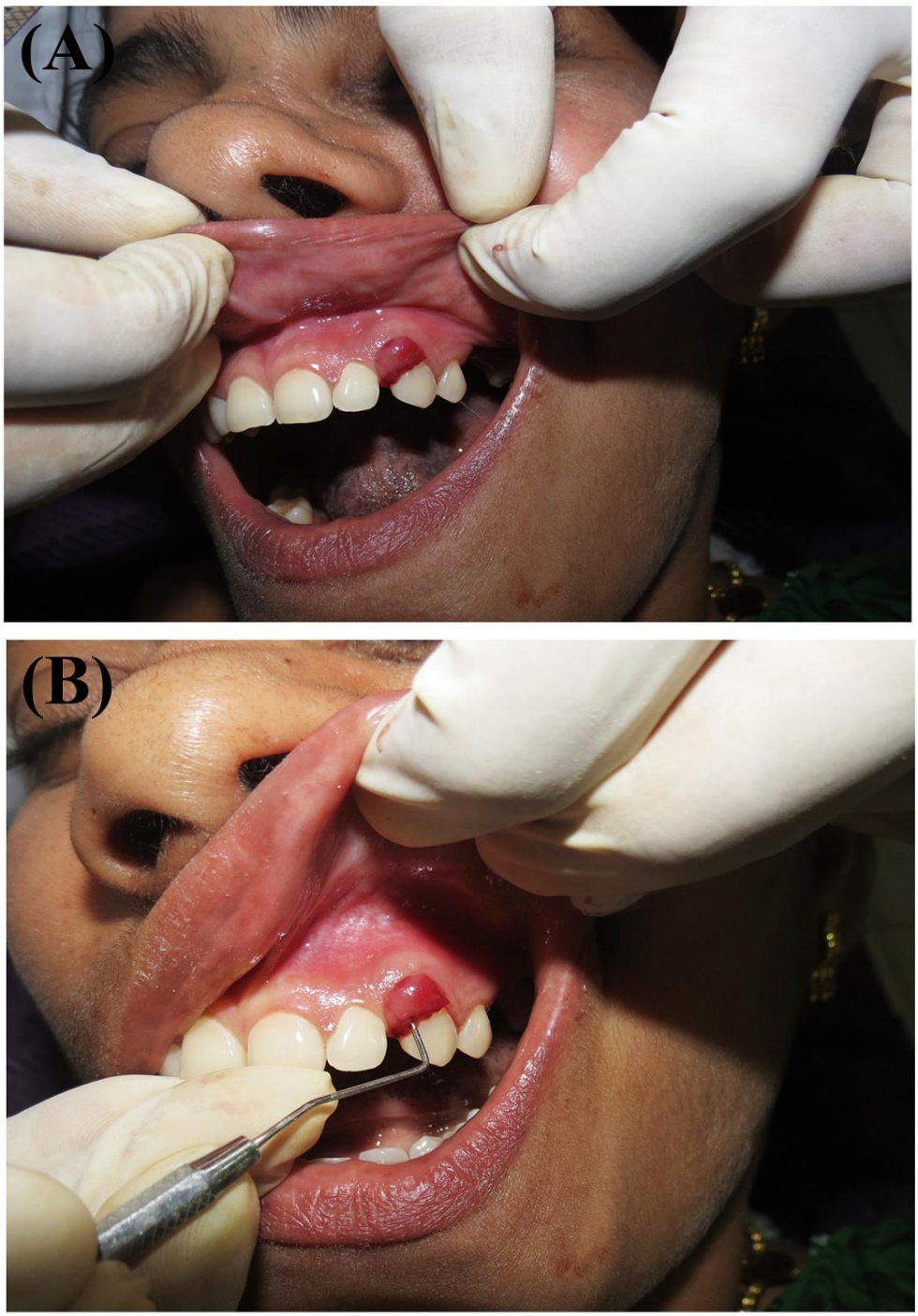


Figure 1: A) Clinical presentation at the time of first visit; **B)** Clinical presentation after three weeks; **C)** Intra-oral radiograph of the same region; **D)** H&E photomicrograph showing plump proliferating endothelial cells in lobular pattern (100X); **E)** H&E photomicrograph showing canalizing channels (100X) and CD 34 immunopositivity (100X, inset); **F)** H&E photomicrograph at 400 X

CASE 2

A twenty four years old female patient reported with a chief complaint of bleeding from gums in the upper left front tooth region since two and a half weeks. On examination, there was an erythematous growth of approximate size 0.8 x 0.9 cm on the attached gingiva in relation to 23 (Fig 2 A). There was a deep pocket of 9 mm (Fig 2 B). The lesion was soft in consistency and was slightly compressible. It bled on provocation. A provisional diagnosis of pyogenic granuloma was made. A modified Widman flap was raised and the lesion was excised and sent for histopathological examination. The submitted section showed tissue covered by para-keratinized stratified squamous epithelium which was atrophied over the connective tissue papillae filled with large dilated lymphatic vessels containing proteic fluid content (Fig. 2 C). The supporting stroma was fibro-cellular. The confirmatory diagnosis was lymphangioma. The lesion did not recur on a 3 month follow up.



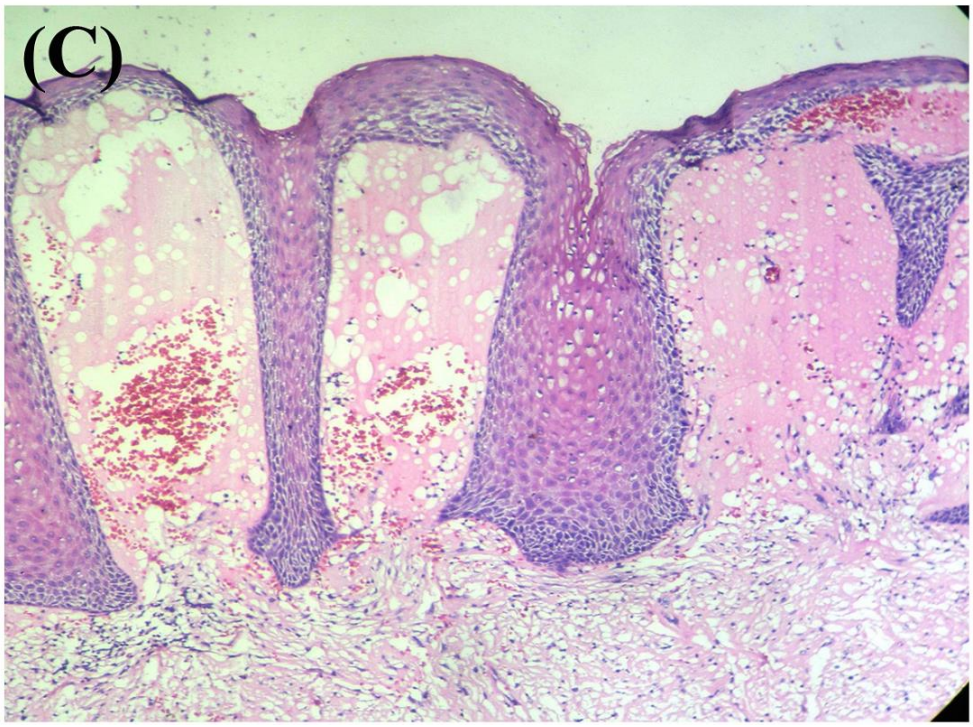


Figure 2: A) and **B)** Clinical presentation in the second quadrant associated with 23; **C)** H&E photomicrograph showing large lymphatic vessels containing eosinophilic homogeneous proteic material filling the connective tissue papillae.

DISCUSSION

Vascular neoplasms including hemangioma and lymphangioma are the infrequent causes of gingival enlargement. It would not be an exaggeration to consider this spectrum of neoplasms as a rare cause of such enlargements on gingiva. We have presented two cases here which clinically resembled a common oral condition, pyogenic granuloma. Both the lesions clinically presented with a non tender erythematous primary growth of gingiva which bled even on slight provocation. But the lesions did not regress with the primary oral health measures and were finally managed by surgical excision. The confirmatory diagnoses were cellular hemangioma and lymphangioma respectively for case 1 and case 2. Routine histopathology and adjuvant immunohistochemistry are thus important for diagnosing such cases.

Hemangiomas may be defined as reddish, bluish or purplish soft vascular lesion which blanches on pressure.[6] In children hemangiomas in children are usually hamartomatous (benign lesions of developmental origin) and involute over time. In contrary to this the adults hemangiomas are usually vascular anomalies and rarely involute spontaneously; rather they typically slowly enlarge. Most common on the tongue, buccal mucosa or lip, as painless reddish, bluish or purplish soft lesions and are usually considered as rare lesion on the gingiva. Most hemangiomas need no treatment but larger should be excised surgically and submitted for histopathological examination.

Oral lymphangiomas are uncommon. Many are of similar structure to hemangiomas but they contain lymph rather than blood. Lymphangiomas are usually solitary and affect the tongue predominantly and gingival lymphangiomas akin to hemangiomas are rare event. Their incidence is approximated to be 1 in 2000 to 4000 live birth. Lesions are classified as macrocystic (single or multiple cysts >2 cm^3), microcystic (<2 cm3), or mixed. The etiology of lymphatic malformations (LM) is unclear. Although most are congenital, there have been reports of LM occurring after trauma or infection. Receptors involved in the formation of lymphatic vascular channels, such as VEGFR3 and Prox-1, may play a role in the development of this disease.[7]

Clinically, the appearance of macrocystic LMs differs from that of microcystic.[7s] Macrocystic LMs present as a soft, fluid-filled swelling beneath normal or slightly discolored skin. Unlike macrocystic LMs, microcystic LMs are soft and non-compressible masses with an overlying area of small vesicles involving the skin or mucosa. These vesicles can weep and at times cause pain or minor bleeding. There is no universal consensus about the clinical nomenclature of oral lymphangiomas. But homologous to skin LMs, we preferred to classify our second case as microcystic lymphangioma. The lesion in the second case was soft mass of size less

than 2 cm^3. But it did not show superficial vesicles. Surgical excision is the treatment of choice. Lymphangiomas do not respond to sclerosing agents as do hemangiomas.[8]

Diagnosis of oral hemangiomas requires some form of imaging to determine their extent and flow characteristics. Ultrasonography can be used to determine that a lesion is angiomatous in nature (i.e., hemangioma, lymphangioma). Furthermore, oral hemangiomas can be differentiated from lymphangiomas by contrast-enhanced MRI.[9] Doppler ultrasound has proven to be a valuable diagnostic method providing images superior to magnetic resonance in cases of microcystic malformations.[10]

Both the cases in our series were clinically diagnosed as pyogenic granuloma and excised in toto. This is because of the fact that gingival lymphangiomas and hemangiomas are extremely rare. The correct diagnosis could be made only after histological examination. Histologically, capillary hemangioma mimics pyogenic granuloma but the former lacks any inflammatory reaction as seen in later aiding in diagnosis. Immunohistochemical markers are employed as adjunctive diagnostic methods which also serve to differentiate hemangioma from lymphangioma. CD34 (positive in case 1 of our series), a surface glycoprotein, is evinced as a blood vascular endothelia marker, whereas lymphatic endothelial cells do not normally express this marker.

In conclusion, vascular neoplasms such as lymphangioma and hemangioma are uncommon rather rare causes of gingival enlargement which may clinically resemble pyogenic granuloma; but do not respond to non surgical treatment. Thus, it is always wise to include these lesions in the differential diagnosis of gingival enlargements.

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