Recurrent Gingival Swelling in Pregnant Women

The aim of this case report is to present a rare case which has been reported with the history of small nodule like growth on mandibular buccal and lingual area. Thirty year old pregnant women reported a nodule on left mandibular buccal and lingual region. The lesion was surgically excised and sent for biopsy to differentiate from kimura's disease, pyogenic granuloma, epithelioid angiosarcoma (EH). Histologically lesion shows proliferation of endothelial cells along blood vessels suggesting epithelioid hemangioma. The purpose of this report is to discuss and differentiate EH and other vascular rare entities occurring in the oral cavity.

KEYWORDS: Epithelioid hemangioma, hemangioma, Kimura's disease, pyogenic granuloma

INTRODUCTION

The hemangioma is vasoformative tumour, characterized by an excess of blood vessels especially veins and capillaries in a focal area of submucosal connective tissue. Oral hemangioma represents 14% of all human hemangioma. There are various subtypes of hemangioma like capillary hemangioma, lobular hemangioma, cellular hemangioma and epithelioid hemangioma (EH).

EH is an uncommon benign vascular lesion which is composed of well-formed immature vessels. It was first described by Wells and Whimster in 1969.[1] The term EH was proposed by Enzinger and Weiss in 1983.[2] The etiology and pathogenesis of this vascular entity is still uncertain.

EH typically arises on the head and neck, but most frequently affected intraoral sites are lips,[3] buccal mucosa and tongue. The extra-cutaneous sites are bone, salivary gland and muscular area or extremities.[4] This entity typically presents as a small angioma like nodule, red to brown in color and may be located intradermally or subcutaneously in young adults. EH can mimic lymphoproliferative disorders, especially when the lesion arises in atypical location such as the extremities.[5] The purpose of this case report is to describe a rare association between EH and oral mucosa.

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CASE REPORT

A thirty year old pregnant (third trimester) female reported to the department, with painless mobile nodular swelling in lower left buccal and lingual area [Figure 1]. There was slight bleeding on brushing and difficulty in speaking. After clinical examination a solitary exophytic growth measuring 1 cm × 2 cm × 1.5 cm was present on mandibular buccal and lingual gingiva in relation to 33 since last one month. Growth was soft in consistency and non tender on palpation. There was no associated regional lymphadenopathy. Radiographic findings showed no overt alveolar bone destruction in the area of soft tissue enlargement. Blood investigation revealed increase in the level of ESR. Blood count did not show eosinophilia. The growth was surgically excised and send for histological and immunofluorescence examination. After three months patient reported back with recurrent gingival swelling in left mandibular canine. The gingiva was soft and tender. This time swelling was decreased in size, and surgically removed.

Microscopic examination

The exophytic overgrowth was excisionally removed and send for histiological examination. The biopsy sections revealed focally ulcerated squamous epithelium. Underlying proliferation of variably sized blood vessel was seen which were lined by plump endothelial cells.

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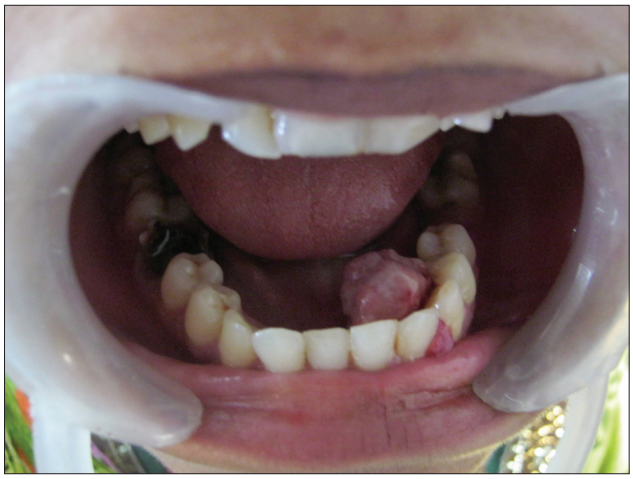

Figure 1: Clinical picture showing exophytic growth on lingual surface of mandibular lower left canine

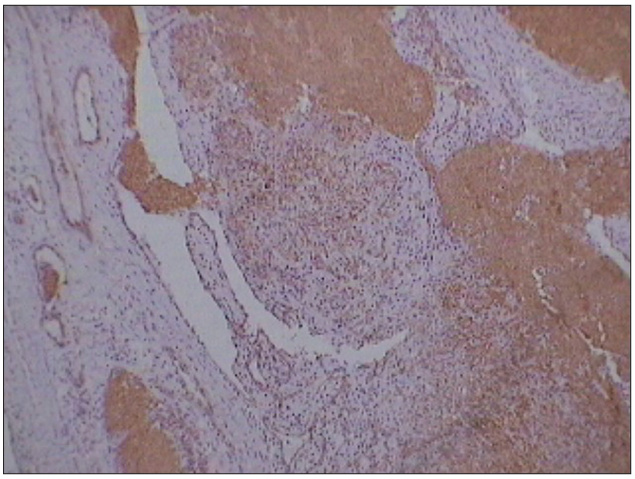

Figure 2: A photo micrograph showing variably sized blood vessel which were lined by plump endothelial cells

Some markedly dilated spaces contained thrombi. The endothelial cells showed round nuclei and eosinophilic cytoplasm. A dense cellular inflammatory infiltrate, mainly lymphocytes, plasma cells and eosinophils and mast cells were found, surroundings the vessels. There were no signs of mitosis and atypical cells [Figure 2].

Immunohistochemistry

It was done using the CD31 marker and revealed the positivity of the cells for CD31 proving that the epithelioid cells seen are vascular in origin.

DISCUSSION

EH is a superficial, often multifocal lesion has a strong predilection for the head and neck region (specially face, ear and scalp) but rare in oral mucosa. EH in the oral mucosa is a rare disease and often confused with other vascular disease. In the past various terms have been used to describe this lesion, including angioblastic hyperplasia with eosinophilia, nodular angioblastic lymphoid hyperplasia with eosinophilia, lymphofolliculosis, pseudopyogenic granuloma, atypical pyogenic granuloma. In 1982, a broad term EH was given to describe angiolymphoid hyperplasia with eosinophilia (ALHE) and some other vascular diseases.[6] But, the diagnosis of epithlioid hemangioma is based upon its histopathological and clinical findings.

Oral EH seems to have a predilection for men, mostly at young age (male: female = 23:13). The lips are the most frequent site (16 out of 36), followed by the tongue (10 out of 36), buccal mucosa (6 out of 36), and palate (3 out of 36).[7] Only 21 cases of EH have been reported in the oral cavity and the gingiva is the most common site.[8] One case has been reported by Misselevich *et al*. which affect alveolar mucosa and gingiva by the direct extension from lesion in mucobuccal fold.[9]

Oral EH clinically present as a solitary asymptomatic nodule, plaques, ulcers or even tumours. Histologically, the vessels were lined by epithelioid or histiocytoid endothelial cells which extend considerably into the lumen, imparting a "tombstone" effect. The lesional cells had rounded nuclei and abundant eosinophilic cytoplasm with occasional vacuoles. Electron microscopy revealed excess mitochondria, endoplasmic reticulum and cytofilaments in the cytoplasm. A mixed chronic inflammatory cell infiltrate, including eosinophils and occasional germinal centers, was seen.[10] CD31 marker were used to check vascular origin of epithelioid cells by immunofluorescence.

The etiology and pathogenesis of EH is unknown. There is a dilemma between various authors whether EH is a reactive lesion or a true neoplastic growth. According to martin – granizor, microtrauma can be a cause of EH. Upto to 10% of cutaneous EH had a history of microtrauma.[11] It is possible that reactive condition caused by trauma may trigger cellular proliferation and growth. Pregnant women have a predilection for EH as reported by Shafer's.[12] Pregnancy along with trauma may exaggerated the cause of exophitic growth as reported in our case. Other hypothesis regarding etiology is infection, allergy, trauma, overgrowth of atypical endothelial cells and inflammatory skin manifestations. Some authors suggested that EH is a true benign neoplasm which has oestrogen receptors on the lesion and respond to oestrogen therapy very well.[13]

EH is differentially diagnosed from other vascular diseases histologically and macroscopically. Macroscopically, EH is differentiated from Kimura's disease, salivary gland tumour, squamous cell carcinoma, lymphoma and pyogenic granuloma whereas histological findings are different in bacillary angiomatosis, epithelioid

angiosarcoma and epithelioid hemangioendothelioma. In Kimura's diseases, prominent cellular areas of lymphocyte forming follicles are seen, which are surrounded by inflammatory infiltrate and fibrosis.[14,15] Bacillary angiomatosis is an infectious disease caused by Rochalimeae henselae, presented as a lesion with abundant neutrophils with nuclear dust and clumps of bacteria.[16] Epithelioid angiosarcoma presents as an infiltrative destructive growth with markedly pleomorphic cells. Cells show atypical mitosis and necrosis.[17] Epithelioid hemangioendothelioma is composed of short cord of spindle shaped endothelial cells. In this lesion well defined vascular channels are not seen.[18]

The line of treatment of oral EH is complete surgical excision and follow-up. Other treatment modalities for cutaneous EH are laser cauterization, diathermy, cryotherapy, retinoids, pentoxifyline and intralesional corticosteroids. Local recurrence is common in one third of patients with cutaneous EH but rarely seen in oral mucosa and treated with re-excision.[16]

CONCLUSION

EH in oral mucosa is rare entity and has very less prevalence rate. The disease requires proper clinical and histopathological examination. As epithelioid hemanigioma is solitary in nature surgical excision is the treatment modality but in case of multifocal involvement, re-excision is required.

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