Case Report

International Journal of Dental and Health Sciences
Volume 05, Issue 02

EPITHELOID HEMANGIOENDOTHELIOMA DISGUISED AS PYOGENIC GRANULOMA: A CASE REPORT

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ABSTRACT:

Epitheloid hemangioendothelioma is a rare tumor with intermediate potency between hemangiomas and angiosarcoma. Most commonly arises from peripheral veins, generally in the lower limb. Intra oral hemangioendothelioma are very rare with only 30 cases published till date. Here we are presenting a case of intra oral epitheloid hemangioendothelioma in a 50 years old male patient appearing as a pedunculated mass on the right side of the maxillary anterior gingiva.

Key Words: Epitheloid hemangioendothelioma, Gingival swelling, vascular tumors of intermediate malignancy

INTRODUCTION:

The term hemangioendothelioma (HE) was first proposed by Borrmann in 1899 with Weiss and Enzinger documented first case in 1982.[1] It is an uncommon vascular neoplasm with intermediate malignant potential between haemangioma and conventional angiosarcoma, associated with high recurrence rate but less chance of distance metastasis.

It can occur at any age but most commonly seen in second to third decade of life with a female predilection, most common sites being peripheral veins of the lower limb, especially from

The iliac and femoral veins, it may also develop in bones, brain, lungs and lymphnodes.[3,4]

kaposiform, Dabska-retiform, compound or complex, low grade polymorphic and epithelioid are the histological subtypes of this tumor with the epitheloid being the most aggressive form.[5]

Clinically the oral epitheloid henangioendotheliomas mimics certain reactive lesions like the pyogenic granuloma present as a bleeding soft tissue growth in most of the cases.[6] In the article we report a case of oral EHE along with a brief review.

CASE DETAIL:

A 50 years old male patient reported with a growth on upper right front region of jaw since three months.it was asymptomatic, He had a habit of Tobacco chewing since last 20years (8 to 10 packets per day) with occasional alcohol consumption.

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On examination the growth was pedunculated, pink in color, size 3x3 cm, smooth texture and soft in consistency with no palpable lymphnodes. It was present on the right side of the maxillary anterior gingiva Anterio-posteriorly extending from mesial side of maxillary right central incisor to mesial side maxillary right 1st premolar, Superio-inferiorly extending from mucogingival junction covering crown of teeth, having lobulated and irregular surface. Radiograph showed no sign of bone resorption and the area appeared to be completely normal.

Based on the clinical examination a provisional of pyogenic granuloma was made.

Under local anaesthesia an excisional biopsy was performed and the specimen was submitted for histopathological examination.

Microscopic examination revealed a highly vascular connective tissue stroma. Within the connective tissue an exuberant proliferation of endothelial cells were seen originating from the dilated blood vessels. The endothelial cells arranged in the form of cords and nests were round to oval in shape with a vesicular nuclei and intracytoplasmic vacuole/lumen formation. Within the lumen erythrocytes were also evident. These epithelial like endothelial cells (epitheloid) were scattered in a myxohyaline stroma. Few mitotic figures were also evident. Intermixed with the tumor cells inflammatory cells (acute and chronic) were also seen.

Later immunohistochemical staining was also done which showed a strong positivity for CD31 and CD34 typically around the vascular channels, thus confirming the endothelial nature of the epithelioid cells. Additionally a negative staining was obtained for cytokeratins, thereby indicating a non- epithelial origin of the tumor cells.

Based on microscopic examination and immunohistochemical findings a confirmed diagnosis of epithelioid haemangioendothelioma was given.

DISCUSSION:

EHE is a rare neoplasm of the vascular endothelium and the first case in the oral cavity was reported by Ellis and Kratochvil in 1986.[7] The etiological factor behind the development of this tumor is unknown yet some authors suggest involvement of T(1; 3)(p36.3; q25), and 22qll. [8]

As described earlier this neoplasm mimics certain reactive lesions due to its intra oral clinical appearance, in our case also the provisional diagnosis was made as pyogenic granuloma and differential diagnosis of peripheral giant cell granuloma, and vascular malformation.

The tumor is characterized by neoplastic proliferation of epithelioid or histiocytic endothelial cells. Predominant locations are soft tissues of extremities, the liver and lungs, involment of the head and neck region is a rare.[9] But our case was found in oral cavity. The most common site in the oral cavity is the maxillary gingiva,

followed by mandibular gingiva and tongue, which is true in our case also.

They can occur at any age with a female predilection, but rare in children,[10] in our case it was found on a male patient.

The different histological variants of HE are epithelioid, retiform, kaposiform, Dabska tumor (papillary intralymphatic angioendothelioma), pseudomyogenic, composite, infantile hepatic, cutaneous, endovascular papillary, histiocytic, mammary, polymorphous, pulmonary, and spindle cell varieties. Epithelioid variant is among the most common types.

Histopathologically, the intraoral epithelioid HE shows cords, strands, solid aggregates of round, oval and polygonal cells, with abundant pale eosinophilic cytoplasm, vesicular nuclei, inconspicuous nucleoli in fibromyxoid or sclerotic stroma with neoplastic cells showing prominent cytoplasmic vacuolization. In most of the cases, the tumor shows no mitotic activity.[11] In our case some mitotic activity was found.

The immunohistochemical markers which can be used in diagnosis of this tumor are CD31, CD34, Ulex europaeus antigen, factor VIII specific antigen to confirm the vascular phenotype of this tumor,[12] vimentin positivity shows the mesenchymal origin of the tumor, KI-67 positivity denotes the increase cellular proliferation of the tumor.[13] In our case also the tumor was positive for CD31, and CD34.

In treatment prospective wide local excision is preferable because of its local recurrence, distant metastasis, and malignant potential.[14, 15]

CONCLUSION:

We conclude that though intraoral epithelioid hemangioendothelioma is rare, the clinician should be aware of clinical and histopathological features of this lesion. Due to its greater rate of malignancy transformation and regional lymph nodes metastasis, treatment mode should be carefully selected.

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FIGURES:

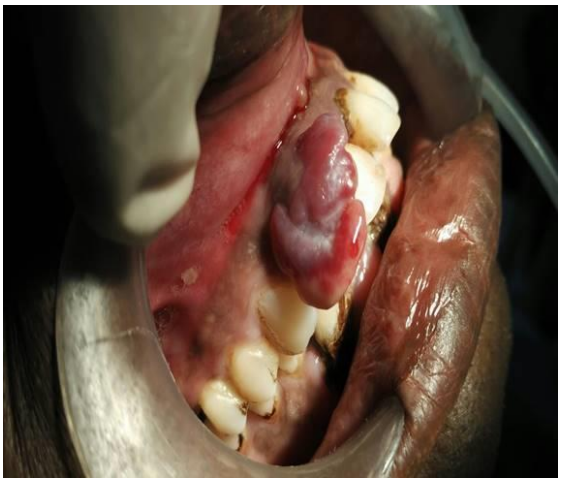

Figure 1

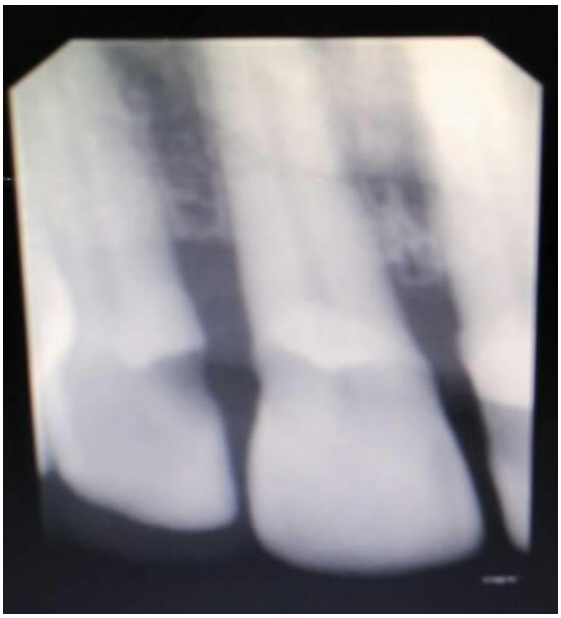

Figure 2

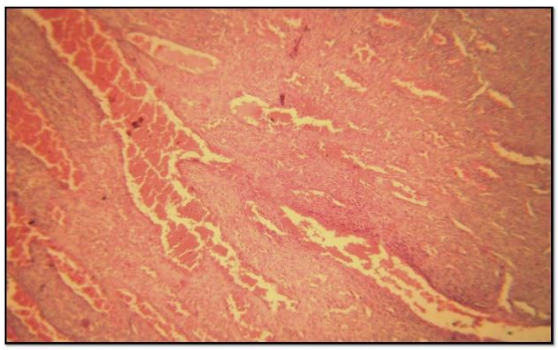

Figure 3

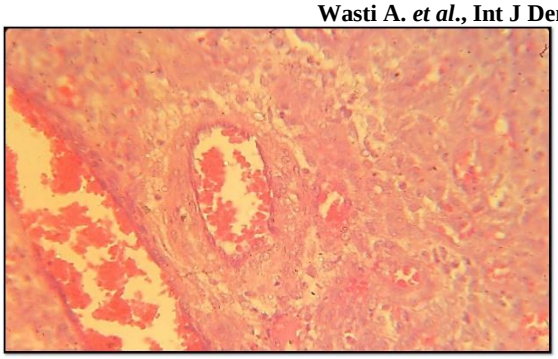

Figure 4

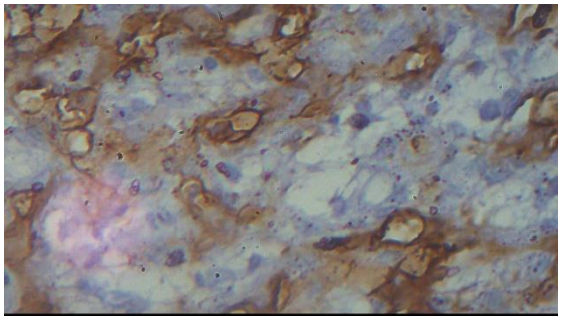

Figure 5