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CASE REPORT

SURGICAL MANAGEMENT OF HEMANGIOPERICYTOMA/SOLITARY FIBROUS TUMOUR OF BUCCAL MUCOSA- A CASE REPORT

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ABSTRACT

Hemangiopericytomas (HPCs)/Solitary fibrous tumors are rare neoplasms of vascular origin that occur in head-and-neck region. These tumors arise from capillary pericytes and are difficult to distinguish from other tumors of vascular origin. HPC, initially described by Stout and Murray in 1942, usually occur in the fifth decade of life and account for 3%–5% of all soft-tissue sarcomas and 1% of all vascular tumors. The tumors usually occur in limbs, pelvis, or head-and-neck region; 15%–30% of all HPCs occur in head and neck. This is a report of a case of HPC located in the right buccal area of a 51-year-old woman.

KEYWORDS: Capillary pericytes,hemangiopericytoma, Immunohistochemistry, vascular tumor

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INTRODUCTION: -

Solitary fibrous tumour is an unusual vascular neoplasm of oral cavity which is not clinically distinguishable from other lesions. First described by Stout & Murray in 1942[1], they are tumours of pericytes of Zimmermann, which are baroreceptors on luminal wall of vascular channels[8]. Pericytes are mesenchymal in origin and are relatively undifferentiated in the sense that they can apparently develop into several different cell types including smooth muscle. They are contractile and can change their shape, thereby reducing the diameter of the capillary lumen[4]. It usually occurs in fourth to fifth decade of life. STFs may show many growth patterns and therefore can be easily mistaken for other more common H&N spindle cell or epithelial lesions[10]. Haemangiopericytoma (HPC) has over the years been used fairly loosely, to describe a wide variety of neoplasms which have certain morphological characteristics in common: a monotonous appearance at low-power examination, moderate to high cellularity, and the presence of numerous, variably thick-walled, branching 'staghorn' vessels[5]. We report a case of HPC of the right buccal mucosa in a 51-year-old female patient.

CASE REPORT:-

A 51-year-old female presented with gradually enlarging asymptomatic mass in her right buccal mucosa for the past 4 months (Fig 1: (a)). The asymptomatic lesion was initially small which gradually enlarged to the present size of 5 cm × 4 cm , which was solitary & well-circumscribed (Fig 1: (b)). The overlying mucosa appeared smooth & mildly erythematous with no visible ulcerations or pulsations. Blanching was seen on the postero inferior aspect of swelling due to impingement of 48. The mass was rubbery in consistency. No lymphadenopathy. The examination of other sub sites of oral cavity was normal.

She had no compounding medical history or any recent history of trauma to the offending site. CT guided incisional biopsy reported it to be hemangio endothelioma. Immunohistochemistry for the same revealed strongly positive CD34.

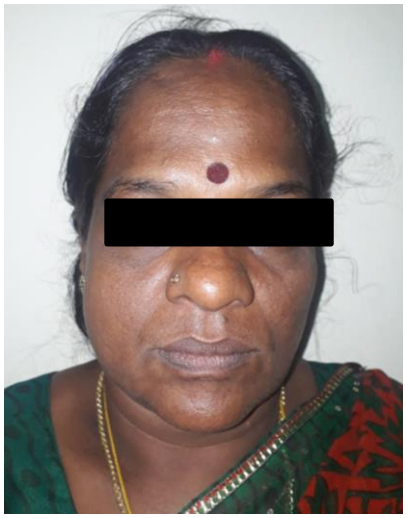

Fig 1:(a)

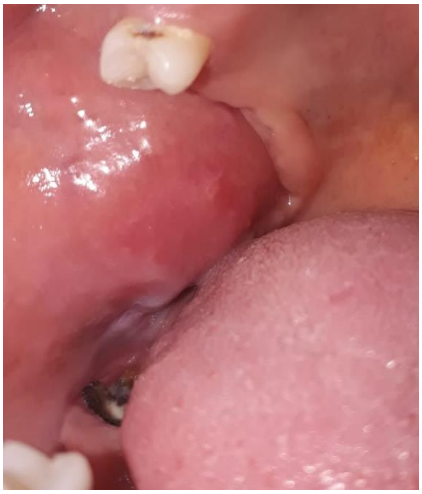

Fig 1: (b)

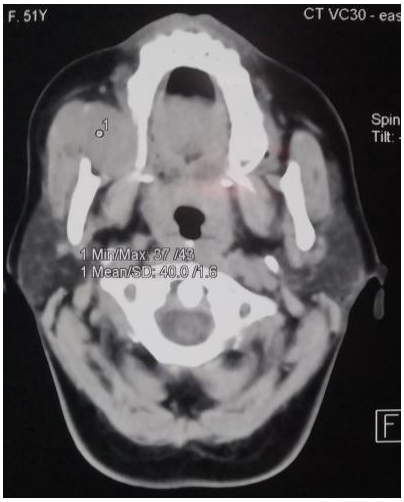

Fig 1:(c)

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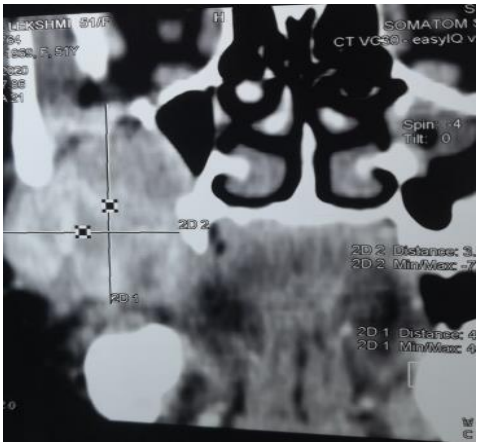

Fig 1: (d)

Fig 1: (a) Extra-oral photograph: an asymptomatic mass in her right cheek (b) Intra-oral photograph: a painless submucosal mass in the right buccal region (c) CT axial cut showing ill-defined heterogenous lesion in right buccal space with infiltration into adjacent masseter, temporalis muscle & pressure erosion of maxilla (d) CT coronal cut showing radiopaque mass in the right buccal space measuring 4.43 X 3.56 cm

TREATMENT AND PROGNOSIS: -

Surgery was conducted under general anaesthesia. A 7.0 cm transcutaneous incision at the right buccal mucosa was made parallel to the anterior border of the mandible ramus. The identified anatomic layers included the mucosa and the buccinator muscle. The tumor was found adjacent to the front part of the buccinator muscle. The tumor was encapsulated with connective tissue. It was easily separated from the layer structure. The tumor was ablated with extracapsular dissection. The patient was discharged 4 days after the surgery. There have been no signs of facial nerve injury or recurrence at 12 months post-operatively. Macroscopically, the cut section of the resected specimen showed a circumscribed erythematous mass measuring 6 ×4 ×2cm surrounded by a fibrous capsule

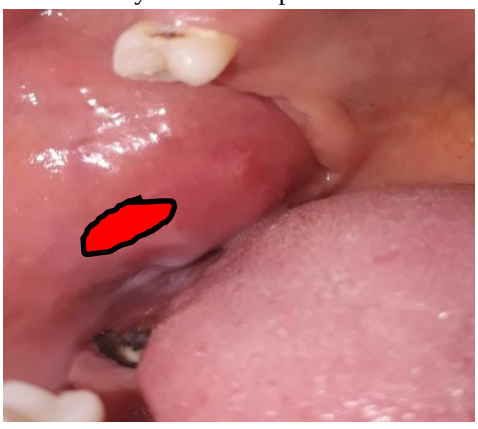

Fig 2:(a)

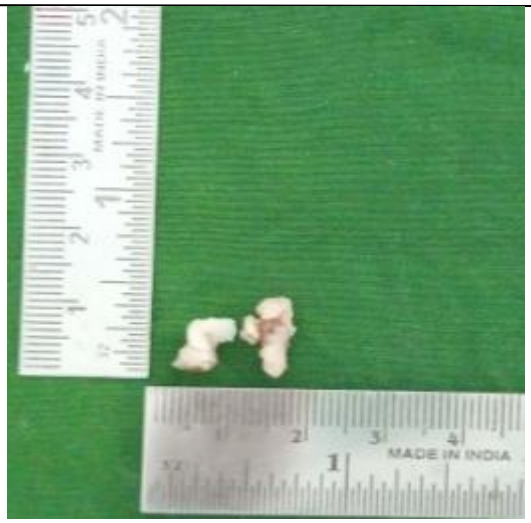

Fig 2: (b)

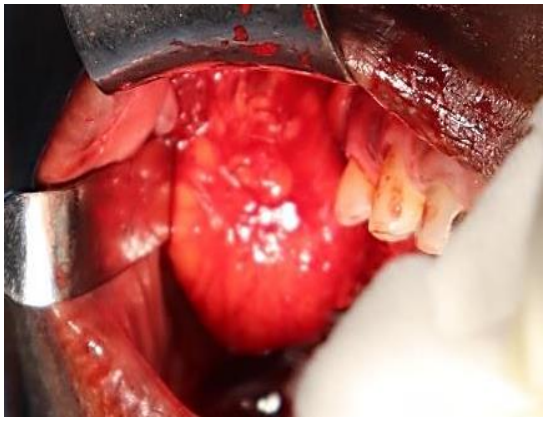

Fig 2:(c)

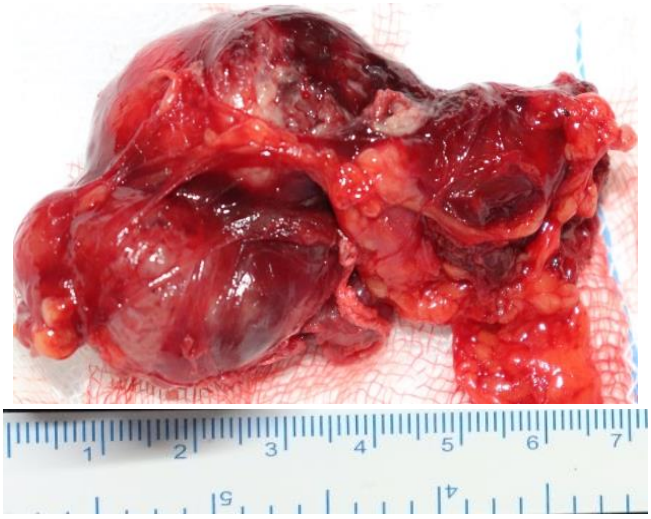

Fig 2: (d)

Fig 2: (a) Incisional biopsy site (b) specimen sent for histopathological examination(c) tumor seen encapsulated with connective tissue (d) Gross appearance of the resected specimen. A well-circumscribed nodular mass measuring 6 ×4 ×2cm surrounded by a fibrous capsule

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HISTOPATHOLOGIC FINDINGS: -

Excised tissue sent for histopathological examination. Histopathology report revealed the presence of neoplasm composed of spindle cells with thin ectactic vascular channels in between. The cells have moderate amount of clear vacuolated cytoplasm & plump mild to moderate pleomorphic nuclei. Mitosis 2-3/hpf in areas. Stroma shows areas of sclerosis. The tumor was labelled as benign HPC because of its of cellular atypia and presence of mitotic activity. The morphological and immunohistochemical features were consistent with the diagnosis of solitary fibrous tumor/hemangiopericytoma.

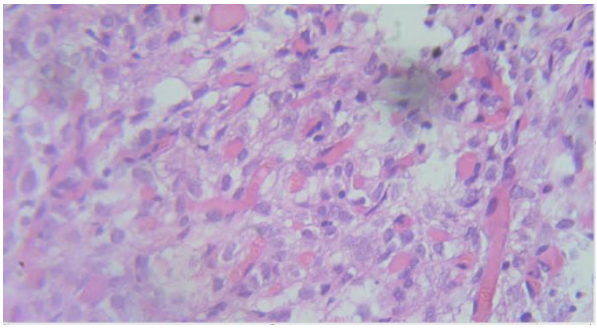

Fig 3:(a)

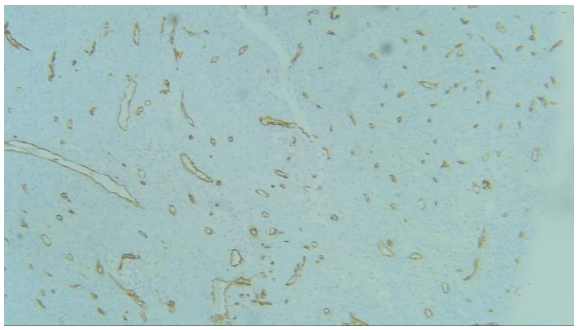

Fig 3: (b)

Fig 3:(a) The tumor comprised spindle cells with an irregular disposition associated with collagen bands and vascular structures branched with an evident lumen (b) Tumor cells showed strong positivity for CD34 (IHC).

DISCUSSION: -

Hemangiopericytomas are benign neoplasms with malignant potential. The sinonasal tract and orbit were the most common sites involved (30% and 25%), followed by the oral cavity and salivary glands (15% and 14%)[7]. They are distinguished based on occurrence of metastasis and recurrence. The appearance on a plain radiograph is not specific. Usually, it presents as radiopaque soft tissue mass. Calcification may occur, but is uncommon. A CT scan and magnetic resonance imaging may aid in identification of the lesion. Angiograms may show evidence of rapid circulation indicated by a richly vascular mass with dilation of the arteries and a diffuse capillary blush or opacification in the arterial phase and dilation of the draining veins in the vicinity of the tumor in the venous phase[2]. Recent IHC evidences now suggest that conceptually this tumor is not derived from the pericyte because it does not express actin or myofibroblastic markers. There is considerable histologic overlap between myofibroma, solitary fibrous tumor, synovial sarcoma, and mesenchymal chondrosarcoma. That is why, the diagnosis of HPC is the diagnosis of exclusion. Chan et al[19] have summarized the essential diagnostic criteria for the diagnosis of SFT which was augmented by strong immunohistochemical CD34 positivity which is a consistent finding in most SFT's confirming the diagnosis. Malignant SFT's are very rare and account to 10–15% of all cases in pleura while in oral cavity there are only two reported cases. Furthermore, there are no unifying histologic criteria that appear to reliably and consistently predict malignancy in SFT. The management of HPC involves wide surgical excision. In the head and neck, cervical lymphadenectomy is reserved for those instances where palpable lymphadenopathy is coexistent. The role of radiotherapy has been questioned because these tumors are generally radioresistant. One study showed that only 13% of patients were cured with radiotherapy. The recurrence rate of SFT occurring in the pleura is reported to be approx. 30%. In contrast, recurrence of an SFT in an oral lesion is rare[13]. More importantly, clinicians should keep in mind that a patient with a past history of SFT can show a malignant relapse even when the pathological features indicated a benign SFT in the first diagnosis. Thus, a continuous long term follow-up is needed for SFT patients.

CONCLUSION

Despite the rarity of HPC, it can be considered as one of the differential diagnosis of tumors of the head and neck region. Though surgical excision is the most accepted management of HPC. Early diagnosis reduces the postsurgical morbidity. Since, local recurrences are very common and late distant metastasis has reported, long-term follow-up, is mandatory both clinically and radiologically.

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Nil

CONFLICT OF INTEREST:

There are no conflicts of interest.

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