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An Unusual Presentation of Fibrosarcoma of Mandible: A Case Report

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Abstract

Soft tissue sarcomas are rare in the head and neck region and account for less than 1% of the malignant tumors. At one time, fibrosarcoma was the most common soft tissue sarcoma. With the advent of electron microscopy and immunohistochemistry, it became evident that many previously diagnosed fibrosarcomas were other spindle cell tumor leading to a reduction in the occurrence percentage of fibrosarcoma. Here we report a rare case of fibrosarcoma presenting as a innocuous growth in the gingiva.

Keywords: Fibrosarcoma, Mandible.

Introduction

Fibrosarcoma is a malignant neoplasm of the fibroblastic origin. Fibrosarcoma of head and neck area represents 5% of all malignant intraosseous tumors. Of all the fibrosarcomas occurring in human, only 0.05% occurs in the head and neck region. Of this, 23% of head and neck fibrosarcoma occur within oral cavity, mandible being the most common site.[1]The exact cause of fibrosarcomas not entirely understood; however, studies indicated that genetic alterations may play a role.

Hyperplastic growths are more common in the oral cavity than neoplastic lesions. The oral environment could be a source of chronic irritation contributing to the development of these conditions. The diagnosis of chronic fibrous hyperplasia is pretty straightforward considering the history, clinical features and appearance. However the possibility of an aggressive lesion should be considered until proven otherwise by histopathological evaluation. This article aims to emphasize this point.

Case history

A 30 year old female patient reported to the department of Oral Medicine and Radiology with the chief complaint of intra oral growth in the left side of lower jaw, posterior region for the past one year. Associated pain was gradual in onset, moderate in intensity, intermittent in nature, pricking type, non-radiating, aggravates on touch and relieves after few minutes. Patient consulted a private institution 4 months back, where incisional biopsy was done and histopathological features gave an impression of fibroma. Patient gives history of quiding betel leaf, nut, slaked lime and tobacco for the past one year with a frequency of 7 times per day. The quid was placed either on the right or left buccal vestibule for a duration of about half an hour and then spat out. Her medical history was non contributory.

Extra oral examination did not reveal any swelling or change of contour. On Intra oral soft tissue examination, a single well-defined exophytic growth was present in the buccal marginal and attached gingiva in relation to 34,35 and 36 of size 3x1.5cm approx, extending supero-inferiorly from marginal gingiva to the buccal vestibule and mesio-distally from mesial aspect of 34 to distal aspect of 36. It was predominantly pale pink in colour interspersed with erythematous areas, roughly oval in shape with smooth lobulated surface, firm in consistency and tender. Intra oral hard tissue examination revealed, Grade III mobility of 35, Grade II mobility of 34 and Grade I mobility of 36.

Intra oral periapical view in the region of 34,35,36 and 37 taken by bisecting angle technique and OPG revealed bone destruction with loss of Periodontal ligament space and lamina dura in 34, 35 and mesial aspect of 36.

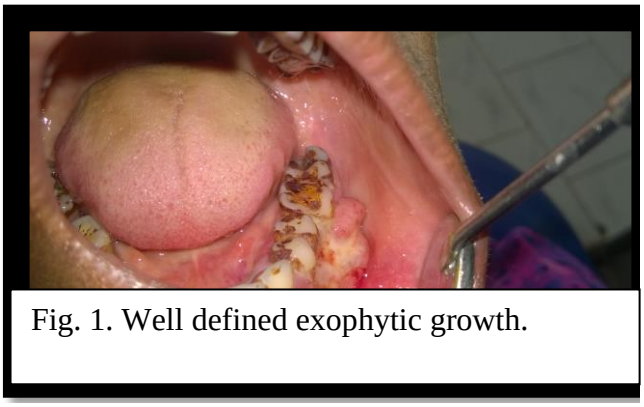

Fig. 1. Well defined exophytic growth.

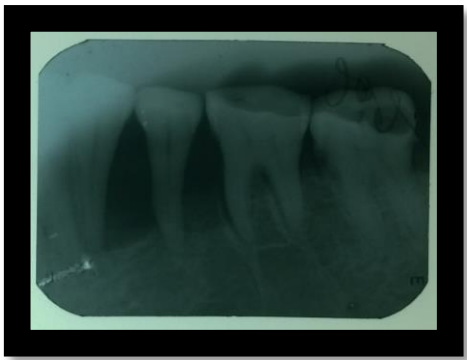

Fig. 2. Intra Oral Periapical Radiograph showing vertical bone loss.

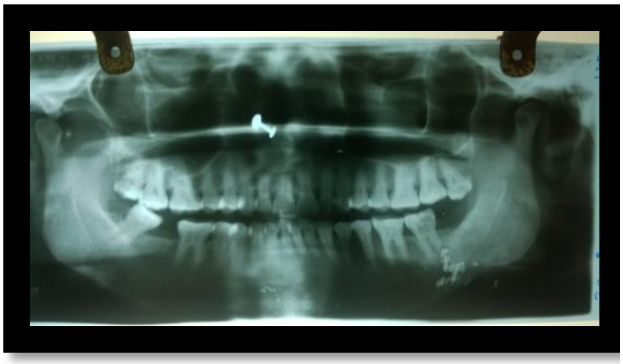

Fig. 3. OPG showing vertical bone loss

An incisional biopsy was performed under local anesthesia. Histopathological examination revealed two bits of tissue. One bit shows parakeratinised stratified squamous epithelium with acanthotic and flat epithelium-connective tissue interface. The underlying connective tissue is densely fibrous and shows an excretory duct, focal vascular proliferation and lymphocyte infiltration. Deeper connective tissue shows interlacing fascicles of spindle cells with large hyperchromatic spindle nuclei.Numerous mitotic figures are noted. The second bit shows interlacing fascicles of hyperchromatic spindle cells. One area show epithelium with mild to moderate epithelial dysplasia of small epithelial islands in the lesional area suggestive of Malignant spindle cell tumour.

Immunohistochemical(IHC) panel of markers were used to identify the lesion among diverse spindle cell malignant tumors. IHC analysis showed cytoplasmic positivity for vimentin, but negative for pancytokeratin, S-100, desmin, muscle specific actin, CD34. Thus by correlating the clinical, histopathological and IHC analysis a final diagnosis of fibrosarcoma was made. Patient was referred to higher centre for further management.

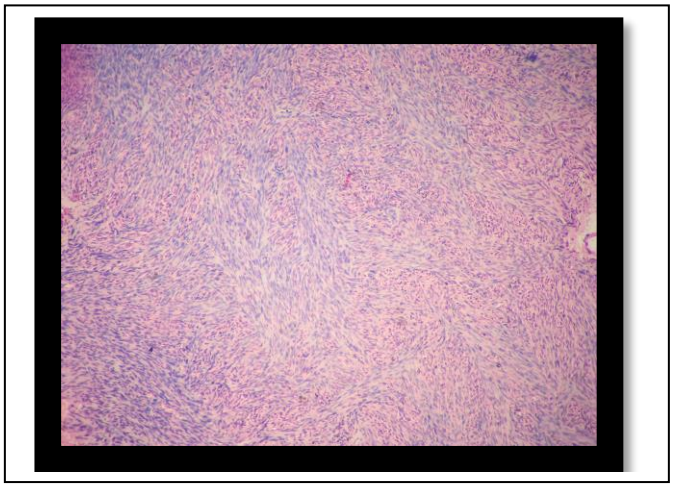

Fig.4 Histopathological picture showing Fibrosarcoma.

Discussion

Fibrosarcoma is a malignant neoplasm of the fibroblastic origin. Fibrosarcoma is defined as a malignant spindle cell tumor showing a herringbone or interlacing fascicular pattern without expression of other connective tissue cell markers.

Fibrosarcoma can arise in soft tissues or within bone. Intraosseous fibrosarcomas may develop endosteally or possibly periosteally. Phemister[2] suggested that this occurs more frequently in the medullary region than in the periosteum.

Dahlin[3] and Schajowicz[4] stated that most fibrosarcoma occur de novo but may arise secondarily from Paget's disease, Ameloblastic fibroma, bone damaged by radiation, chronic osteomyelitis, rarely fibrous dysplsia; it may also occur as a malignant transformation of giant-cell tumor of the bone.

Fibrosarcoma in the head and neck region develop in the 3rd and 5th decade of life, but there is a wide age range and many patients are below 20 years of age. Infantile or congenital fibrosarcoma is the most common soft tissue sarcoma found in children under 1 year of age. [5-7] There is no gender predilection. However, in some studies male predilection has been reported. Any submucosal site may be involved although buccal mucosa, tongue and alveolus accounts for more than half of the cases.

The symptoms of fibrosarcoma vary depending on size, location and spread of the tumor. Fibrosarcoma of the oral cavity most often manifests as a clinically innocuous, lobulated, sessile, painless and nonhemorrhagic submucosal mass of normal coloration. Pain and paresthesia are usually late symptoms indicating nerve involvement. On the other hand, aggressive fibrosarcoma tend to be rapidly enlarging, hemorrhagic mass similar in clinical appearance to an ulcerated pyogenic granuloma, peripheral giant cell granuloma or peripheral ossifying fibroma. Even lesions that do not demonstrate surface ulceration or rapid growth may show destruction of underlying muscle and bone.[7-9]

The mode of spread is via local invasion and haematogenous dissemination. A latent period of 10 years before metastasis appear is quite observed and the commonest site for metastases being the lungs. The most widely used staging

system for fibrosarcomas was developed by the American Joint Committee on Cancer. The categories of this system include grade (G), size of the tumor (T), lymph node involvement (N) and presence of metastases (M). Low grade and high grade are designated G1 and G3, respectively. The size of the tumor can be <5 cm (2 inches), designated as T1, or >5 cm, designated as T2. If the lymph nodes are involved, it is designated N1, while no lymph involvement is designated N0. Finally, there may be a presence of distant metastases (M1) or no metastases (M0). The following is a list of stages and their indications:

Stage IA: G1, T1, N0, M0

Stage IB: G1, T2, N0, M0

Stage IIA: G2, T1, N0, M0

Stage IIB: G2, T2, N0, M0

Stage IIIA: G3, T1, N0, M0

Stage IIIB: G3, T2, N0, M0

Stage IVA: Any G, any T, N1, M0

Stage IVB: Any G, any T, N1, M1

Tumors with lower stage numbers, such as IA and IB, contain cells that look very similar to normal cells, while tumors with higher stage designations are composed of cells that appear very different from normal cells. In higher staged tumors, the cells appear undifferentiated.[9,10]

Radiographically, fibrosarcoma often appears as a purely osteolytic lesion with a geographic, moth eaten, or permeative pattern of bone destruction. Generally a wide zone of transition is evident with no neoplastic new bone formation. The absence of tumoural calcification or ossification can be of importance in differentiating fibrosarcomas from other malignancies such as chondrosarcomas and osteosarcomas. Fibrosarcoma is the only malignant bone tumor in which sequestration is found frequently; sequestration also has been described in the intracortical Ewing's sarcoma.[11]

Histopathologically, fibrosarcoma has been characterized by uniform spindle cells distributed in interlacing fascicles with herring bone growth pattern. The differential diagnosis for fibrosarcomas include all spindle cell tumors, and only careful examinations of multiple sections and special stains as well as immunohistochemical analysis will permit a correct diagnosis.[9]

The treatment of choice is surgical resection with a wide margin. The need for adjuvant radiotherapy and/or chemotherapy is still unclear but there is normally an indication in high grade tumours because these tumours may present with subclinical or microscopic metastases at the time of diagnosis. The need for prophylactic neck dissection is controversially discussed and it is not performed in all cases. The overall survival rate at 10 years may vary from 21.8% to 83%, and clinical stage, histological grade of malignancy, and local recurrences are the most important prognostic factors.[7]

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