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# Benign Fibrous Histiocytoma- An Unusual Case

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## Abstract

Benign fibrous histiocytoma (BFH) is a tumour of the soft tissues of mesenchymal origin. Its main constituents include fibroblasts and histiocytes. This tumour is commonly associated with upper and lower limbs and rarely with bony tissue. It is a rare entity in the maxillofacial region and less than twenty cases have been reported till date. This case describes a 60 year old male with a tumour affecting the mandible. Radiographically, it appeared as a multilocular expansile radiolucency with sclerotic margins associated with a pathological fracture of the right angle of mandible.

**Keywords:** Benign Lesions, Oral And Maxillofacial Surgery, Oral And Maxillofacial Pathology, Rare Benign Lesions, Histiocytoma, Fibrous Histiocytoma, Benign Fibrous Histiocytoma.

## Introduction

Fibrous histiocytoma (FH) was first described by Stout & Lattes in 1967. [1] It is a tumour of the soft tissues arising from the mesenchymal cells and is mainly made of fibroblasts and histiocytes arranged in a cartwheel or storiform pattern, along with a varying number of inflammatory cells, foam cells & siderophages. [2] It is commonly observed in the skin of the upper and lower extremities and bones like pelvis, femur and tibia. Involvement of either the maxilla or the mandible with this pathology is rare. This tumour has been called by various names like dermatofibroma, histiocytoma cutis, nodular superficial fibrosis, sclerosing hemangioma, xanthogranuloma & fibroxanthoma.[3,4] This tumour has the ability to convert into its malignant variant called " Malignant Fibrous Histiocytoma" (MFH) or "Pleomorphic Sarcoma". [5] In the oral cavity, it is associated with buccal mucosa, gingiva, lips, soft palate & floor of the mouth and the upper and lower jaws and rarely involved. [1]

## Case Report

A 60 year old male patient reported to the Department Of Oral And Maxillofacial Surgery complaining of swelling on his right lower jaw region since 2 months. The patient stated that initially the swelling had been small and then it slowly grew to its present size. The swelling was

associated with localised mild, intermittent, dull, aching, non-radiating pain with no aggravating and relieving factors. There was also an associated tingling sensation at the chin region. He had no history of episodes of fever, pus discharge or any other associated sign and symptoms related to the swelling. Other relevant history included his adverse habit of smoking 10-12 beedis a day since 25-30 years but had quit the habit 3-4 years back. Extra-orally, the swelling was approximately 3cm× 2cm and confined to the right lower jaw region. Antero-posteriorly, it extended from the right commissure of the lip to a point 2cm anterior to the right earlobe and from the right commissure of the lip to the inferior border of the mandible supero-inferiorly. The swelling on palpation was bony hard and fluctuant in some areas, suggestive of cortical perforation and the overlying skin appeared normal. On palpation of the lower border of the mandible, a step and associated tenderness was observed at the right angle region. A solitary right submandibular lymph node of 1cm × 1cm was palpable. It was firm, mobile and non-tender in nature. Intra orally, the lesion extended from the distal aspect of right second premolar(45) to the angle of the mandible with vestibular obliteration. The right first and second molars (46 and 47) were mobile. The overlying mucosa was normal and swelling showed similar palpatory findings as those on extra oral examination. (Figure 1,2, 3)

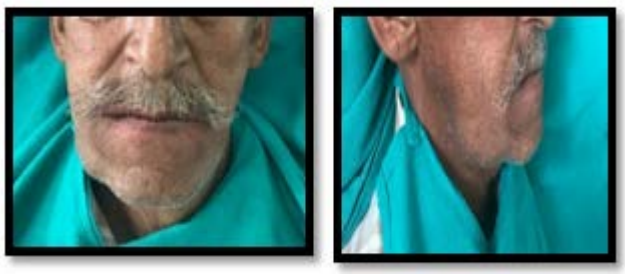

Figure 1 And 2: Extra Oral Extent of Lesion

Figure 3: Intra Oral Extent of Lesion With Healing Socket Of 47 Post Extraction

The Orthopantomogram (OPG) revealed the presence of a large, expansile, multilocular swelling extending from the subapical region of 46, involving the apical region of 47 upto the angle of the mandible. A breach in the continuity of the inferior border of the mandible was present below 47 region suggestive of a pathological fracture. (Figure 4) The Cone Beam Computed Tomography (CBCT) showed a hyperdense lesion extending from 46 to the right ascending ramus of mandible with buccal expansion, thinning of buccal and lingual cortical plates and involvement of the right inferior alveolar nerve. The lesion had a size of 3.6 cm supero-inferiorly, 2.4 cm antero-posteriorly and 29.5mm medio-laterally. There was a breach in the continuity of the inferior border of the mandible just below 47 indicating a pathological fracture.(Figure 5) Based on the abovementioned findings, our provisional diagnoses included osteomyelitis and ossifying fibroma. Aspiration was attempted and it was negative. An open biopsy was planned under local anaesthesia with extraction of 46 and 47. On histopathological examination, fibroblasts and fibrocytes were seen arranged in a storiform pattern. Spindle shaped uninucleated cells with scanty cytoplasm and dense

inflammatory infiltrate was also observed suggestive of a fibrous tumour.

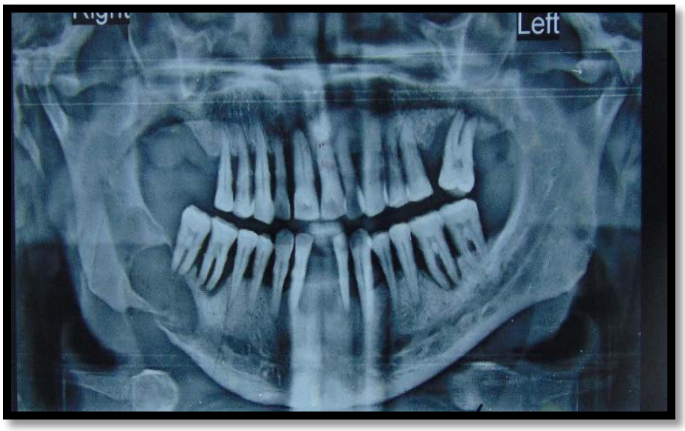


Figure 4: OPG Showing Pathological Fracture and Multi Locular Radiolucency

Figure 5: CBCT Showing Hyperdense Lesion With Involvement Of The Right Inferior Alveolar Nerve.

A 3D Rapid Prototyping model was constructed of the patient's jaw using the CBCT data for a mock surgery. Resection of the pathology was planned followed by placement of a titanium reconstruction plate. The entire procedure was performed on the model save surgical time. (Figure 6 and 7) Arch bars were placed to aid in inter maxillary fixation.(IMF)

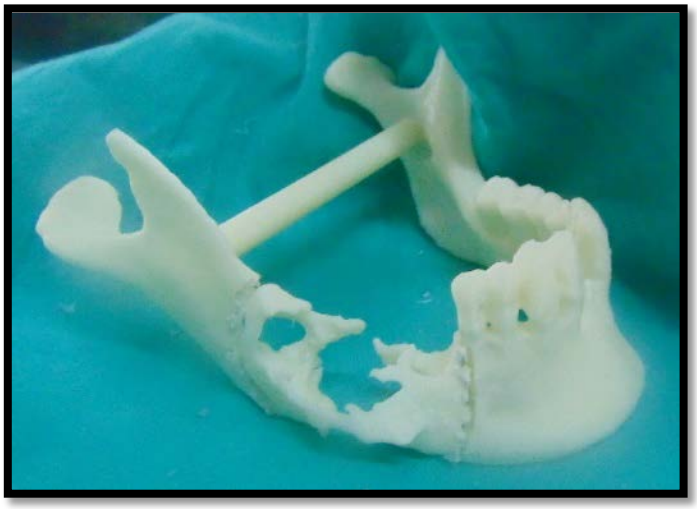

Figure 6: 3D Model with Osteotomy Cuts on Each Side of the Lesion

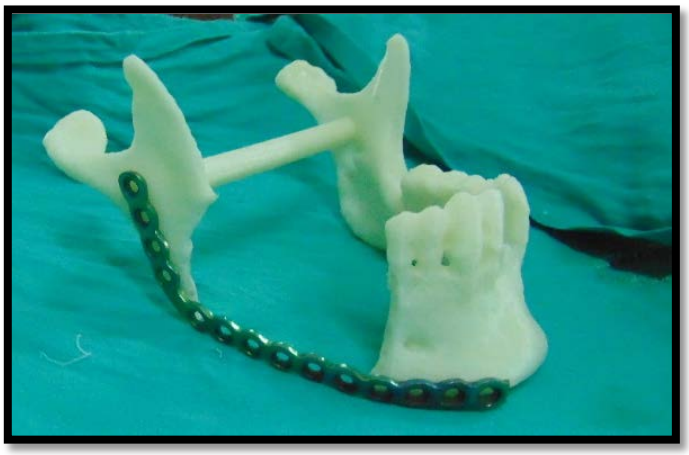

Figure 7: 3D Model with Pre Bent Titanium Plate on The Inferior Border of Mandible

Under General Anesthesia, the tumor was excised using an intra oral approach. patient's surgery was then planned under general anaesthesia using an intra oral approach.(Figure 8) Bony margins were smoothened and the pre bent right angle titanium reconstruction plate was placed using 5 (2.5mm× 10mm) titanium screws.

(Figure 9 and 10) Closure was done in layers. The surgical site healed uneventfully.

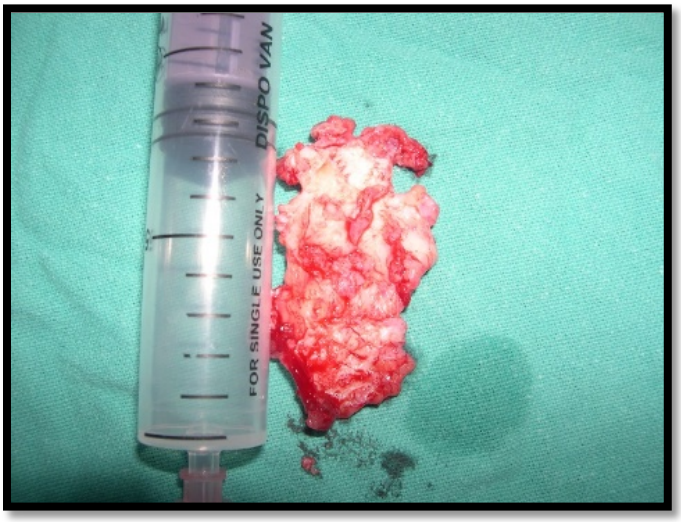


Figure 8: Specimen After Excision

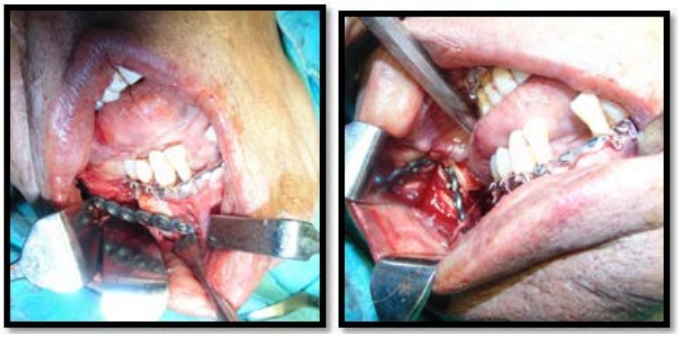

Figure 9 and 10: Reconstruction Plate In Place After Surgical Excision of Lesion.

On histopathological examination of the surgical specimen, the findings were similar as that of open biopsy and the sample was then sent for immunohistochemistry. (IHC) This revealed strong positive markers for CD68 i.e. marker for histiocytes and Vimentin i.e. marker for connective tissue. The specimen was examined for a third marker i.e.CD34 which is a marker of endothelial cells. (Figure 11-14) The result was found to be negative. Based on the IHC and histopathological findings, this tumour was diagnosed as a BFH.

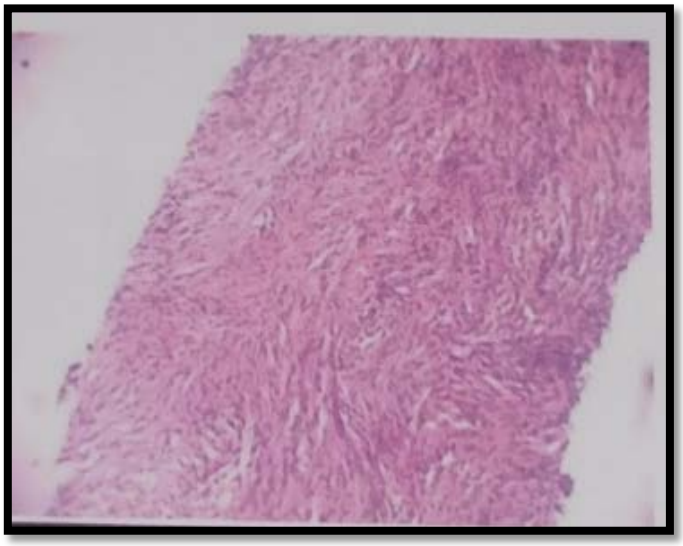

Figure 11: Histopathological Examination

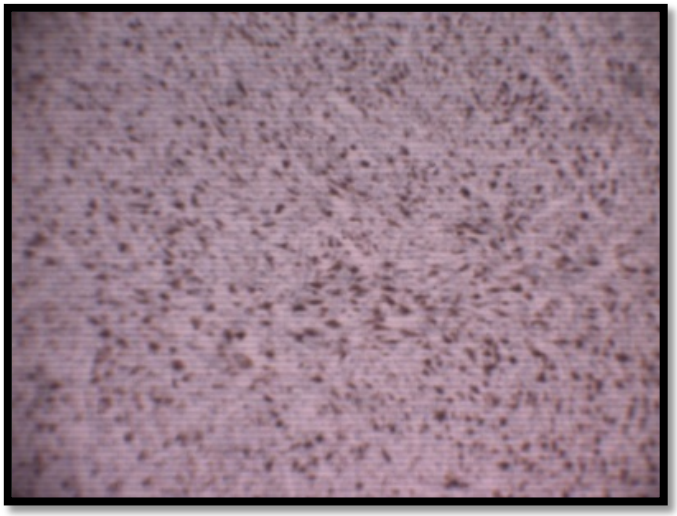

Figure 12: Positive CD68

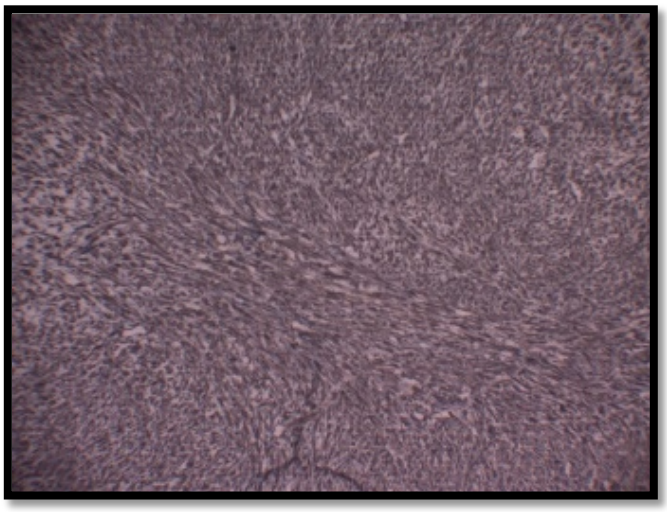

Figure 13: Positive Marker For Vimentin

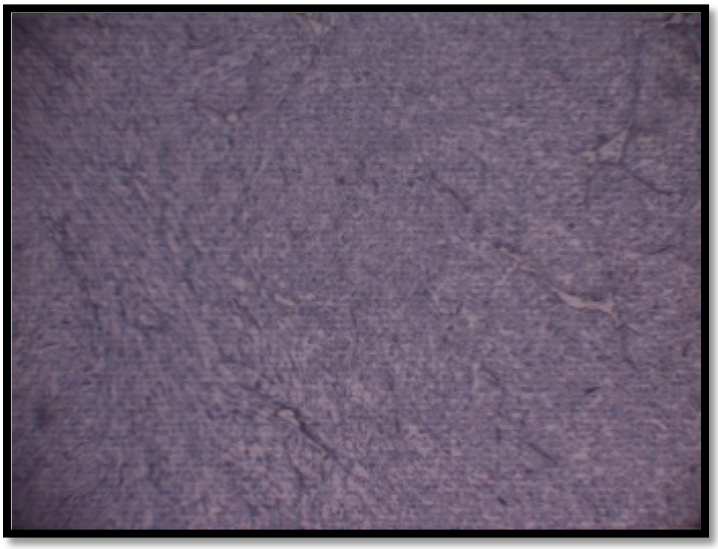

Figure 14: Negative For CD34.

One month post- operatively the patient showed adequate wound healing and complete absence of swelling from the affected site. (Figure 15 and 16)

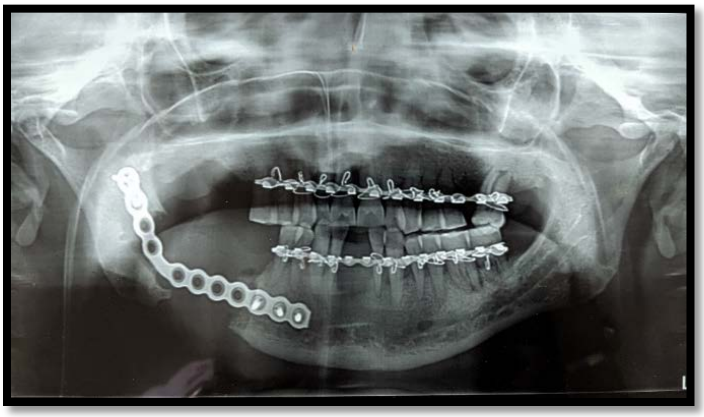

Figure 15: OPG showing adapted reconstruction plate on the mandible.

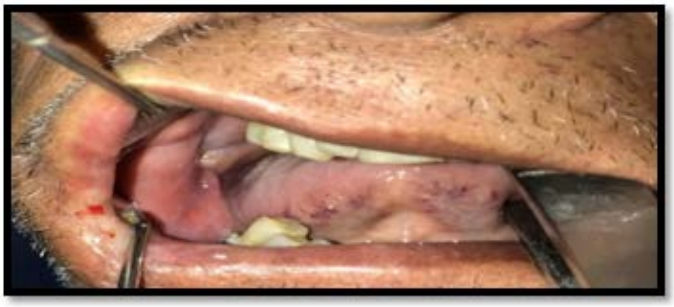

Figure 16: Intra Oral View Of Surgical Site

**Discussion**

FH is used to describe a broad group of proliferative lesions that are usually of benign character. [6] In a more contemporary analysis, Hoffman and Martinez separated these lesions into two broad categories, benign and malignant, and subdivided these into 1) benign fibrous histiocytoma, 2) atypical fibrous histiocytoma, 3) inflammatory fibrous histiocytoma, and 4) malignant fibrous histiocytoma. [7] The common age of occurrence for BFH is usually in the fifth decade with male to female ratio of 2.5:1. The aetiology of oral BFH is obscure. Chronic irritation, continuous trauma & spontaneous development have been suggested as the possible etiological factors.[8] These tumours usually present as slow growing, solitary nodule on the surface of extremities. The oral lesions are typically found in the middle aged & older adults & vary in size from few millimetres to several centimetres. Deeper tumours tend to be larger. They are usually asymptomatic but itching or pain often are noted. [3] Histological variants of BFH include cellular, epitheloid & aneurysmal as three main variants along with clear cell, lipidized, palisading, myxoid & granular cell types. [2] In addition Han et al classified it as fibro-collagenous, histiocytic, cellular, aneurysmal, angiomatous, sclerotic, monster, palisading & keloidal types. Amongst all these cellular variant is found to have most aggressive behaviour & high risk of local recurrence (25%) & often poses a diagnostic challenge to distinguish it from more aggressive lesions such as fibrosarcoma.[9] The differential diagnosis of oral BFH includes MFH, fibrosarcoma, solitary fibrous tumour, angiomatoid fibrous histiocytoma & leiomyoma. [10] Treatment of choice is usually wide surgical resection along with safe margins of 2mm for soft tissue lesions & 5mm for bony lesions.[4] Some authors state that this

lesion reoccurs while others say it has poor chances of recurrence. Either way, a close follow up is essential.

**Conclusion**

BFH presents not only a diagnostic dilemma to the surgeon but has also been called a "pathologist's phantasm." Therefore, thorough clinical examination of this rare lesion is essential for proper treatment plan. Excision of the lesion is recommended with a long term follow up to detect recurrences and presence of malignant findings.

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