Case Report

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Dentistry Section

Benign Fibrous Histiocytoma of Mandible: A Case Report and Updated Review

MANJUSHA PATTAMPARAMBATH[1], SUDHA SATHYABHAMA[2], ROHITASHWA KHATRI[3], SUJATHA VARMA[4], NAVAJEEVRAJ MACHIPALLIYIL NARAYANAN[5]

ABSTRACT

Benign Fibrous Histiocytoma (BFH) is a well recognised tumour of the soft tissue, developing entirely within the subcutaneous tissue, the deep soft tissues or in the parenchymal organs. However, BFH of bones is poorly defined. It has been rarely reported in the bones with femur, tibia and pelvic bone being the most commonly affected bones. Involvement of the jawbones is rare. Their clinical and radiographic features often simulate the common odontogenic and non-odontogenic lesions of the jaws. Hence, it is imperative for a dentist to have proper knowledge of various conditions affecting the jaws. Here we report a case of BFH which presented as an indolent swelling of the right mandibular posterior region for 15 years. The case emphasizes the need for considering BFH in the differential diagnosis of swellings of the jaws.

Keywords: Fibroblasic, Histiocytic, Swelling

CASE REPORT

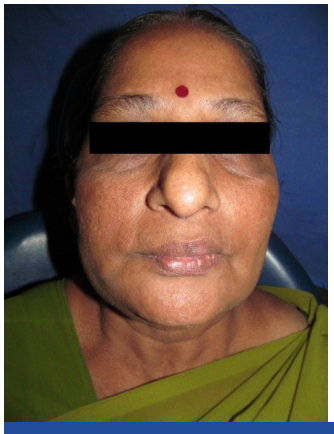
A 51-year-old female patient presented to the Department of Oral Medicine, with an asymptomatic swelling of the right side of lower jaw. Patient noted the swelling 15 years back when a dentist whom she consulted for the treatment of carious teeth drove her attention to it. Carious teeth, 46 and 47 were extracted. Though the patient was advised to undergo a biopsy she didn't return to the dentist. Over the past 15 years the swelling had only slightly increased in size. A month ago, the patient consulted another dentist for extraction of mobile 45, who noticed the swelling and referred the patient to our institution for further evaluation.

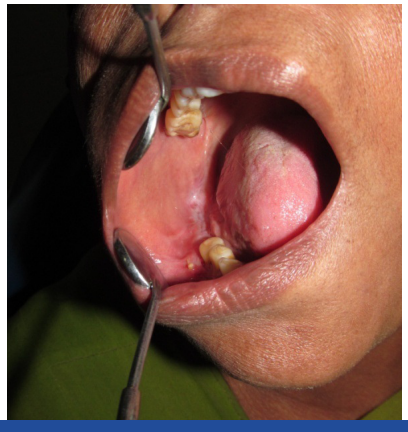
Extra orally a mild facial asymmetry was noticed in the right mandibular body region [Table/Fig-1]. Further examination revealed a diffuse hard non tender swelling of size 5x5cm extending from corner of mouth to 5cm anterior to pinna. Superiorly it extended from level of corner of mouth to the lower border of mandible which was expanded. On palpation borders were ill defined. The swelling had a smooth surface and the skin overlying was normal. There was no history of pain or pus discharge in the associated area or any local rise of temperature. The patient's medical and family history was non contributory, and physical examination revealed no other abnormality. Buccal cortical expansion was noted. Intra orally mucosa was normal, multiple missing teeth were noted in the right quadrant [Table/Fig-2]. Right second premolar was grade II mobile.

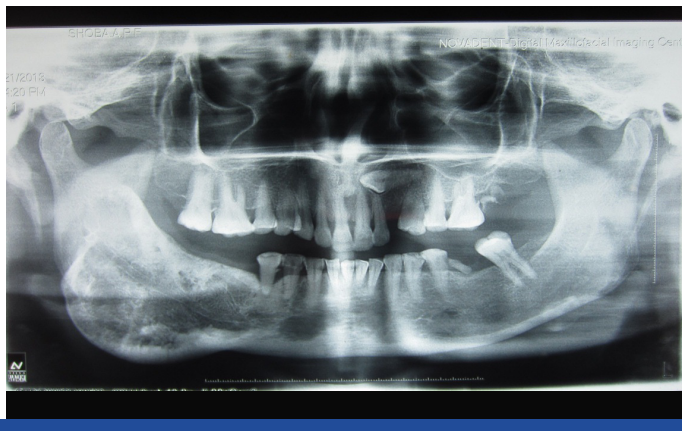
Radiographs were advised. Panoramic view revealed a mixed radioopaque radiolucent lesion with ill defined borders extending from distal aspect of right second premolar to the ramus region posteriorly. Superoinferiorly the swelling extended from the edentulous alveolar crest to the inferior border with the expansion of lower border, angle and ramus of the mandible [Table/Fig-3]. PA view showed a mixed radio opaque radiolucent lesion with buccal cortical expansion [Table/Fig-4]. Occlusal radiograph revealed bicortical expansion [Table/Fig-5]. Computed Tomography (CT) revealed a lytic lesion of size 6x6cm in right mandible with buccal and lingual cortical expansion [Table/Fig-6,7].

Diagnosis: Based on the clinical presentation of painless slow growing swelling in the posterior mandible and the radiographic presentation of ill defined mixed radioopaque radiolucent lesion, odontogenic tumours like calcifying epithelial odontogenic tumour, desmoplastic ameloblastoma and calcifying cystic odontogenic tumour were considered in the differential diagnosis. Other non odontogenic lesions like fibrous dysplasia, ossifying fibroma and osteoblastoma were also considered.

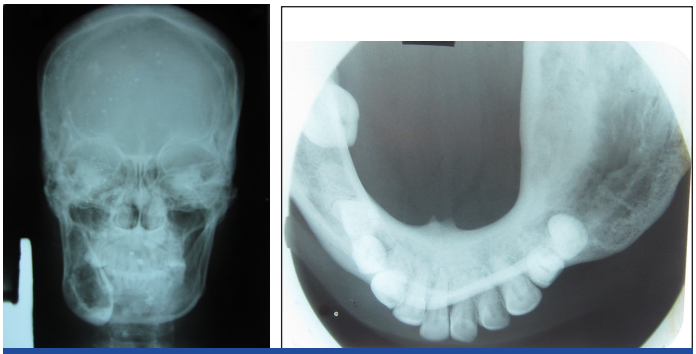
An incisional biopsy was performed under local anaesthesia. Histopathological examination revealed proliferation of tightly packed, spindle-shaped, fibroblast like cells with elongated uniform basophilic nuclei arranged in interlacing bundles, streaming fascicles and in storiform pattern [Table/Fig-8]. Rounded xanthoma like cells (foamy histiocytes) with pale eosinophilic







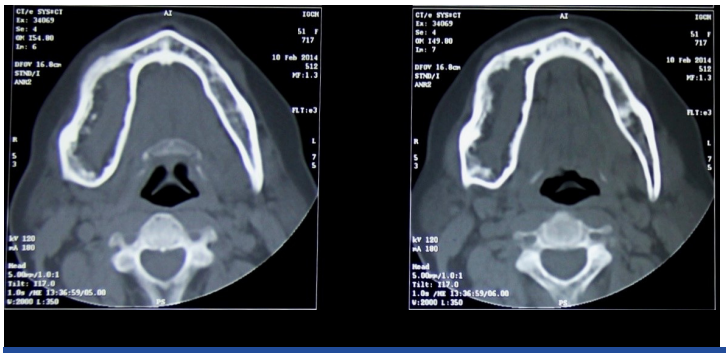
[Table/Fig-1]: Extra oral photograph of the patient showing mild asymmetry of right lower face. **[Table/Fig-2]:** Intra oral view. **[Table/Fig-3]:** Panoramic radiograph showing mixed radio opaque radiolucent lesion with ill defined borders.



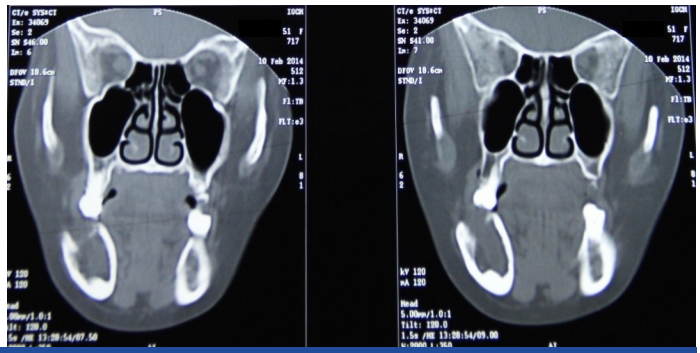
[Table/Fig-4]: Postero-anterior view showing mixed radio opaque radiolucent lesion. **[Table/Fig-5]:** Occlusal radiograph showing buccal and lingual cortical plate expansion.

evidence of nuclear or cellular atypia or mitosis was evident. A provisional diagnosis of benign spindle cell neoplasm was made. Immunohistochemical analysis was performed to confirm the fibrohistiocytic nature of the lesion. The tissue was diffusely positive for vimentin [Table/Fig-11] while focally positive for CD68 [Table/Fig-12]. The positivity for CD68 and vimentin demonstrated that the lesion were composed of histiocytic cells and fibroblast-like cells respectively. The negativity for S-100 differentiated the lesion from neurogenic tumours. A diagnosis of benign fibrous histiocytoma was reached.

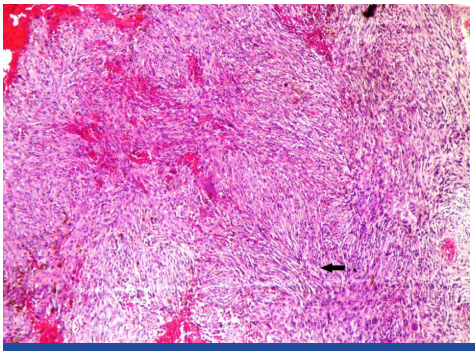
Patient was operated under general anesthesia and segmental mandibulectomy [Table/Fig-13,14] was performed. The microscopical examination of excised specimen revealed similar findings as in incisional biopsy. The patient did not report back for post-op evaluation.

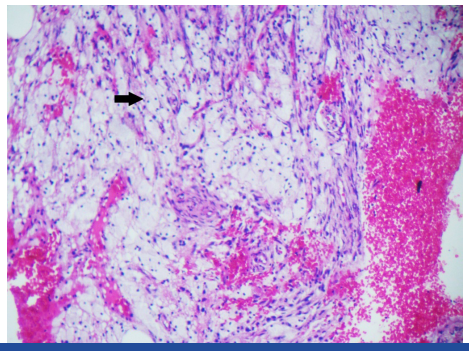


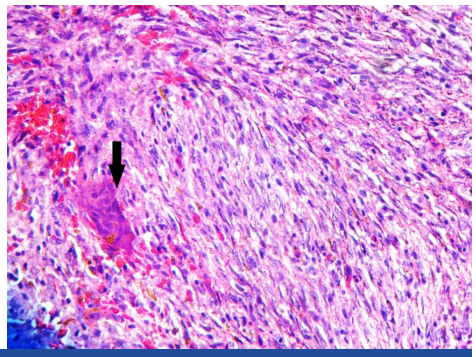
[Table/Fig-6]: CT axial view showing expansile lesion.



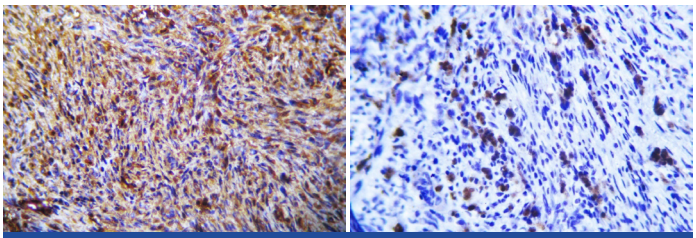
[Table/Fig-7]: CT coronal view.







[Table/Fig-8]: H & E stained section at 100x magnification showing storiform pattern arrangement of spindle shaped cells. **[Table/Fig-9]:** H & E stained section showing sheets of foamy histiocytes and areas of haemorrhage 100x. **[Table/Fig-10]:** H&E stained section at 400x magnification, giant cell seen.



[Table/Fig-11]: Tumour cells diffusely positive for vimentin. **[Table/Fig-12]:** Tumour cells focally positive for CD 68.

[Table/Fig-13]: Intra operative view. **[Table/Fig-14]:** Post operative view.

cytoplasm and eccentrically placed basophilic nucleus were seen scattered within the stroma [Table/Fig-9]. A few multinucleated giant cells, scattered lymphocytes, areas of haemorrhage and haemosiderin pigmentation were also seen [Table/Fig-10]. No

DISCUSSION

Benign Fibrous Histiocytoma (BFH) represents a diverse group of neoplasms which exhibit both fibroblastic and histiocytic differentiation. The term “Benign Fibrous Histiocytoma” was first described by Stout and Lates in 1967 [1]. BFH is a relatively common and well characterized soft tissue lesion with the majority of them affecting the skin of extremities in the early to middle adult life [2,3]. Primary BFH of the bone is rare, approximating to 1% of all benign bone tumors [4]. There are less than 100 reported cases, with femur and tibia most frequently involved [5].

Oral and maxillofacial region is a relatively rare site for BFHs and involvement of jaws is extremely rare [6-12]. Only a few cases of BFH in the mandible have been reported [13]. Patients have ranged in age from 6 to 74 years at the time of diagnosis, 60% being older than age 20 years, with a slight female prevalence [1]. In the current case patient was a female of 51years which is similar to that of earlier reported cases.

Buccolingual expansion with multilocular radiolucency and a sclerotic rim around the osteolytic defect are typical features in cases reported in the posterior region of the mandible [12]. However, in our case the radiographic appearance was that of a mixed radio opaque radiolucent lesion which had an ill defined border.

The basic pattern of BFH consists of a stroma of spindle-shaped fibroblasts, arranged, at least focally, in a whorled, storiform pattern, among which a variable number of small, multinucleated, osteoclast-type giant cells are scattered [5]. Foam (xanthoma) cells, scattered inflammatory cells, mainly lymphocytes, stromal haemorrhages and deposits of haemosiderin pigment may also be present.

Histological findings of BFH are indistinguishable from those of the Fibrous Cortical Defect (FCD) and Non-Ossifying Fibroma (NOF). Hence, they should be differentiated on clinical and radiographic presentations. Fibrous cortical defect and nonossifying fibroma are asymptomatic, self-limiting developmental defects confined to the metaphyseal portion of the long bones in children and adolescents. They are usually discovered as incidental radiographic findings. Fibrous cortical defect appears as a small radiolucent lesion isolated to the cortex of the involved bone, while nonossifying fibroma is slightly larger and involves a portion of or the entire width of the medullary cavity and may cause spontaneous fracture of the involved bone. On the contrary, though BFH lesions may occur at any age, they are more common in adults. The occurrence of BFH is seen in non-long bones and even in case of long bone involvement no metaphyseal involvement has been reported [13].

BFH must be distinguished from fibrohistiocytic degenerative or repair tissue that occurs in other bone lesions, most notably and frequently from central giant cell granuloma, brown tumour of hyperparathyroidism etc. Histopathologically central giant cell granuloma demonstrates multinucleate giant cells in a cellular vascular stroma containing spindle shaped cells. However, the spindle cells lack the typical arrangement seen in BFH. Although brown tumour of hyperparathyroidism shares histopathologic features with central giant cell granuloma, it can be excluded with serum calcium and phosphate determination and parathormone assay. Hypercalcemia, hypophosphatemia, and elevated parathyroid hormone blood levels characterize hyperparathyroidism [14].

Immunohistochemistry acts as a useful aid in diagnosis by confirming the origin of cells. CD68 is a transmembrane glycoprotein that is highly expressed by human monocytes and tissue macrophages. It is particularly useful as a marker for the various cells of the macrophage lineage including monocytes, histiocytes, giant cells, Kupffer cells and osteoclasts. In the current case the positivity for CD68 demonstrate the histiocytic nature of the cells where as vimentin confirms the presence of fibroblast-like cells.

Cale et al., proposed that BFH could be diagnosed in the jaws under 2 circumstances [7]: (1) when the tissue exactly resembled metaphyseal fibrous defects microscopically and clinically the patient was an adult and had pain or swelling with radiologic evidence of a locally destructive radiolucent lesion, most often in the mandibular angle-ramus area or (2) when the jaw lesion exactly revealed the features of the soft tissue BFH on microscopic examination. In the present case the jaw lesion occurred in an adult and presented as a locally destructive lesion in the posterior mandible and the tissue resembled metaphyseal fibrous defects microscopically.

Common modalities of treatment for bony BFH are curettage, complete resection of the tumor and bone grafting. The prognosis of BFH is good and recurrence is rare. However, some of the reported cases have shown a locally aggressive behaviour as in the present case which calls for a prolonged follow-up. The current patient is being followed up regularly and is free of recurrence three years post surgery [Table/Fig-13,14]. Malignant transformation in BFH is extremely rare. However, Tanaka et al., reported a case in mandible which underwent malignant transformation [15].

CONCLUSION

It is imperative for a physician dealing with the head and neck area to have an accurate knowledge of various lesions that may affect the jaws. BFH is a rare entity in the mandible with a few cases reported so far. BFH constitutes a diagnostic dilemma because it shares common clinical symptoms, radiological characteristics, and histological features with a diverse set of lesions, such as odontogenic tumours, nonossifying fibromas, fibrous dysplasia, osteoblastomas, etc. Therefore, a definitive diagnosis of BFH should only be made by evaluating the patient's symptoms, tumor location, radiological characteristics, histopathological features, and immunohistochemical staining results.

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PARTICULARS OF CONTRIBUTORS:
1. Senior Resident, Department of Oral Pathology and Microbiology, Government Dental College, Calicut, Kerela, India.
2. Professor and Head, Department of Oral Pathology and Microbiology, Government Dental College, Calicut, Kerela, India.
3. Senior Resident, Department of Oral Medicine, Government Dental College, Alappuzha, Kerela, India.
4. Assistant Professor, Department of Oral Pathology and Microbiology, Government Dental College, Calicut, Kerela, India.
5. Assistant Professor, Department of Oral Pathology and Microbiology, Government Dental College, Calicut, Kerela, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Dr. Manjusha Pattamparambath,
1/1097-A, Sukrutham, Near Kanakalaya Bank, P.O Westhill, Calicut- 673005, Kerela, India.
E-mail: manjumdvn@gmail.com

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