Case Report

A rare presentation of benign fibrous histiocytoma in the maxilla

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Abstract

Fibrous histiocytoma is a soft-tissue tumor that may present as a fibrous mass anywhere in the human body. The involvement of the oral cavity is rare. We here report a case of benign fibrous histiocytoma localized in the maxilla. A 25-year-old male presented with a slowly increasing large painless mass over the left side of the upper jaw for 5 months. The swelling was of gradual onset, slowly progressive and was not associated with pain, tenderness, or discharge. The radiographic appearance showed a multilocular mixed radiodense, radiolucent lesion in the left posterior maxilla. The histopathological report showed connective tissue component having streaming fascicles of spindle-shaped cells showing a storiform pattern. Many areas showed foamy histiocytes along with few multinucleated giant cells. The cells were appearing benign without any appearance of atypia. The tumor cells were positive for CD-68 and vimentin and negative for CD-34 and S100. The lesion was excised under general anesthesia.

Keywords: Benign fibrous histiocytoma, maxilla, storiform pattern

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INTRODUCTION

Fibrous histiocytic lesion such as benign fibrous histiocytoma (BFH) is a rare entity. BFH of bone is included under a group of lesion termed as "Fibrohistiocytic tumors of bone."[1,2] It was first described by Stout and Lattes in the year 1967 as a soft-tissue neoplasm, most commonly seen in the skin as solitary, slow-growing nodule, which targets the mid-adult life age group. However, the involvement of jaw bone is extremely rare with only two cases of the maxilla has been reported in the literature.[2,3] Here, we report a case of BFH with an unusual central presentation in the left posterior maxilla.

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CASE REPORT

A 24-year-old Hindu male reported to the outpatient department with the chief complaint of painless swelling in the upper left back gum region for 6 months. The swelling was of gradual onset, slowly progressive and was not associated with any pain or discharge. Dental history revealed a similar swelling in the same region for which he had undergone surgical excision of the lesion followed by extraction of the associated tooth and root canal treatment of the adjacent tooth. His medical and family history was noncontributory and general physical examination revealed no other abnormalities. Extraoral examination revealed

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the presence of a diffuse swelling on the left side of the face. On palpation, a solitary swelling was present which was slightly tender and firm inconsistency. Intraorally on inspection, a well-defined swelling of size 3 cm × 2 cm in its greatest dimension was present on the left side of the maxilla, extending from the mesial side of maxillary first premolar to the distal side of maxillary first molar. The mucosa over the swelling appears stretched, shiny and pale with patches of erythematous areas [Figure 1]. There was no evident pus discharge or bleeding seen. Loss of buccal vestibular depth was noted. On palpation, the swelling was slightly tender, firm in consistency, sessile which is nonreducible, noncompressible and nonfluctuant. There was no pus discharge or bleeding or any other associated symptoms on palpation. The involved root canal treated tooth did not show signs of mobility or tenderness. A provisional diagnosis of multicystic ameloblastoma and differential diagnosis of the calcifying epithelial odontogenic tumor, calcifying odontogenic cyst, fibrous dysplasia, ossifying fibroma and osteoblastoma were also considered. Radiographic examination included intraoral periapical radiograph of the upper left posterior region, orthopantomogram, and cone-beam computed tomography. It revealed a well-defined mixed radio-dense, radiolucent lesion having multilocular appearance seen in the posterior aspect of the left maxilla measuring about 40.4 mm anteroposteriorly, 30.7 mm mesiodistally, and 41.4 mm superoinferiorly. Anteriorly, the lesion was extending from the region of premolars until the pterygoid plates [Figure 2]. Superiorly, the lesion was extending up to the level of the middle meatus. There was evidence of buccal cortical plate expansion followed by the perforation of the buccal cortical plate. There was also evidence of root canal treated 26 and root resorption withrespect.to 25, 26. Routine blood as well as biochemical investigations were within the normal limits. Incisional biopsy was advised, and the H&E stained section of the specimen showed a connective tissue component having streaming fascicles of spindle-shaped cells showing storiform pattern [Figure 3a]. Many areas showed foamy histiocytes along with few multinucleated giant cells [Figure 3b and c]. Few areas were fibrous in nature with areas of hyalinization. The cells were appearing benign without any presence of atypia. The histopathological diagnosis of "Benign spindle cell neoplasm" suggestive of "Benign Fibrous Histiocytoma" was given and Immunohistochemistry was advised to confirm the fibrous histiocytic nature of the lesion. The tissue was focally positive for CD 68 and vimentin which confirmed that the lesions were composed of histiocytic- and fibroblast-like cells, respectively [Figure 3d]. The negativity of S100 and CD34 differentiated the lesion from neurogenic and vascular tumors, respectively. This helped us to reach a definitive diagnosis of "Benign fibrous histiocytoma." The lesion was excised under general anesthesia, and the patient has been followed up periodically but no recurrence and other changes have been noted for the past 5 months.

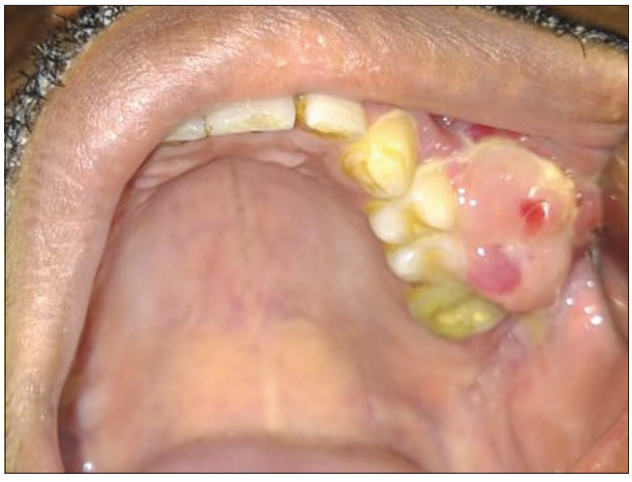

Figure 1: Intra-oral photograph showing well-defined solitary swelling in the left posterior maxilla

DISCUSSION

Fibrous histiocytoma represents a group of neoplastic or quasi-neoplastic lesion which exhibits a mixture of

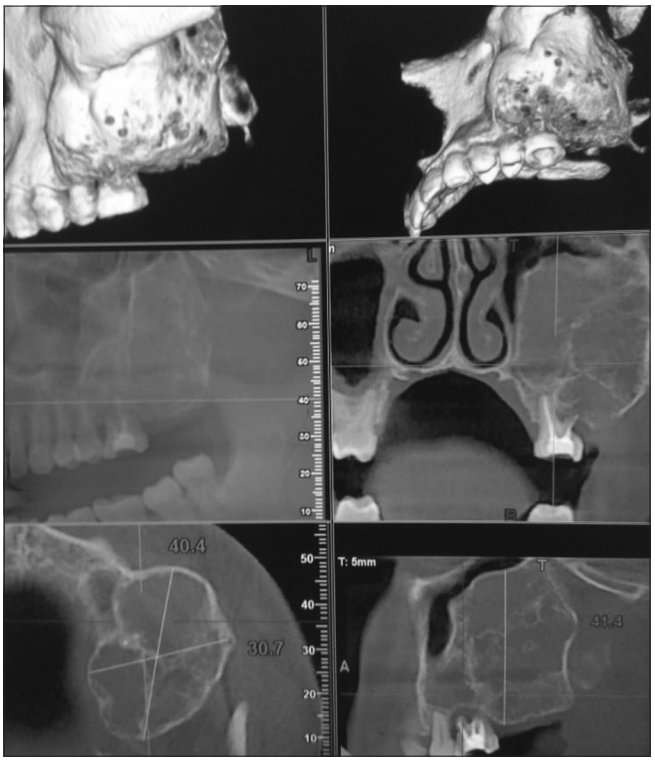

Figure 2: Cone-beam computed tomography revealing mixed radiodense radiolucent lesion in the left maxillary posterior region

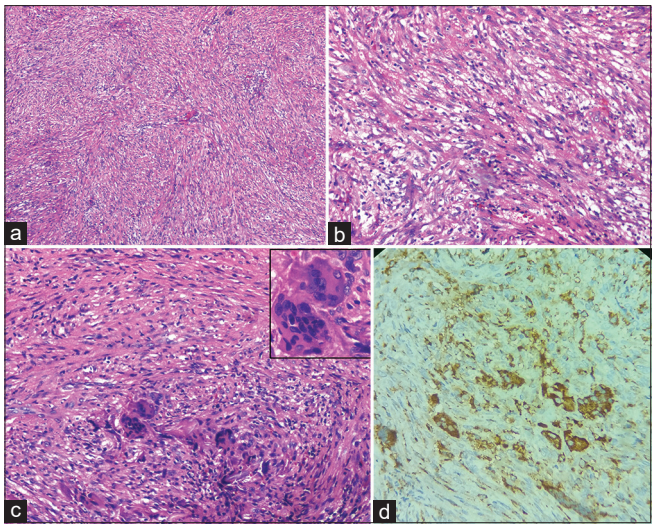

Figure 3: (a) Section showing a storiform pattern (H&E). (b and c) Showing histiocytes, fibroblasts and multinucleated giant cells. (d) immunohistochemical showing CD68-positive cells

fibroblastic and histiocytic cell differentiation.[4] The oral and maxillofacial region is a relatively rare site for BFHs, and the involvement of jaws are extremely rare with <100 reported cases according to the World Health Organization classification of tumors.[5] From a review of literature of BFH involving the oral and maxillofacial region, it is clear that the occurrence of this tumor in the jaw bone is rare as only two cases of the maxilla and seven cases of the mandible have been reported.[4] There is a great dispute among pathologists as to whether this tumor represents a true neoplasm, a reactive process, or a developmental defect. The etiology is not yet clear.[6] Some experts say that the cells originate from tissue histiocytes and then assumes fibroblastic property, while others hypothesize that the positivity of IHC marker factor XIIa favors dermal dendrocytic cell origin.[4] Hence, in the consequence of the controversies of origin, BFH has been designated by various names such as sclerosing hemangioma, hemangioma cutis, fibroxanthoma, and nodular subepidermal fibrosis.[7] The clinical picture as seen in the literature is reported within a mean age group of 40 years with predominance in male adults (2.5:1).[8] According to the reports, BFH has an affinity toward the left side, and the most common site includes the mandibular posterior region followed by the maxillary posterior region and the mandibular anterior region.[6] The present case was reported in a 24-year-old male and in the left posterior maxilla. Clinically, these benign tumors can present as painless, solitary, gradually enlarging, from 2 to 3 cm up to more than 10 cm, over a period of several months that does not show aggressive behavior or damaged overlying mucosa.[8] The current case showed same picture with a well-defined swelling of size 3 cm × 2 cm in its greatest dimension. Other complaints include frequent nasal obstruction, nasal discharge, episodes of epistaxis and ear pain which were not evident in this case.[6,9] Buccolingual expansion with multilocular radiolucency and a sclerotic rim around the osteolytic defect are the characteristic features as reported in the literature.[3] However, in our case, the radiographic appearance was that of a mixed radiodense radiolucent lesion having a multilocular appearance. Histological features of BFH in bones are indistinguishable from those occurring in the soft tissues. Proliferating fibrohistiocytic cells arranged in a storiform pattern is the hallmark of this tumor.[9] Foam (xanthoma) cells, scattered inflammatory cells, mainly lymphocytes, stromal hemorrhage's and deposits of hemosiderin pigment may also be present.[6] Xanthoma cells were not present in the current case. Immunohistochemistry has a vital role in confirming the origin of cells. CD68 is a transmembrane glycoprotein that is highly expressed by human monocytes and tissue macrophages and is used marker for the various cells such as monocytes, histiocytes, giant cells, Kupffer cells and osteoclasts.[3] In the current case, the positivity for CD68 and vimentin confirms the histiocytic and fibroblastic nature of the cells. The prognosis of oral BFH is very good[1,6] as metastases have not been reported till date. However, local recurrence is seen only if there is an incomplete excision of the lesion. Hence, it is necessary that the lesions should have wide margins because simple enucleation of the tumor from the surrounding tissue may facilitate local recurrences.[1] Malignant transformation of BFH is rare. However, Tanaka *et al*., reported a case in mandible which underwent a malignant transformation.[10] Hence, a long-term periodic follow-up is necessary.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Prasanna Kumar D, Umesh, Rathi T, Jain V. Benign fibrous histiocytoma: A rare case report and literature review. J Maxillofac Oral Surg 2016;15:116-20.

2. Cale AE, Freedman PD, Kerpel SM, Lumerman H. Benign fibrous histiocytoma of the maxilla. Oral Surg Oral Med Oral Pathol 1989;68:444-50.
3. Pattamparambath M, Sathyabhama S, Khatri R, Varma S, Narayanan NM. Benign fibrous histiocytoma of mandible: A case report and updated review. J Clin Diagn Res 2016;10:ZD24-6.
4. Prasanna K. Oral benign fibrous histiocytoma – A review of literature from 1964-2016. Adv Dent Oral Health 2018;8:555-748.
5. Kishino M, Murakami S, Toyosawa S, Nakatani A, Ogawa Y, Ishida T, *et al*. Benign fibrous histiocytoma of the mandible. J Oral Pathol Med 2005;34:190-2.
6. Saluja H, Kasat VO, Rudagi BM, Dehane V, Kalburge JV, Nikam A. Benign fibrous histiocytoma of the maxilla: A case report and review of literature. Indian J Dent Res 2014;25:115-8.
7. Neville BW, Damm DD, Allen CM, Chi A. Oral and Maxillofacial Pathology. 1st ed. South Asian: Elsevier; 2015. p. 479-80.
8. Menditti D, Laino L, Mezzogiorno A, Sava S, Bianchi A, Caruso G, *et al*. Oral benign fibrous histiocytoma: Two case reports. Cases J 2009;2:9343.
9. Shoor H, Pai KM, Shergill AK, Kamath AT. Benign fibrous histiocytoma: A rare case involving jaw bone. Contemp Clin Dent 2015;6:S266-8.
10. Tanaka T, Kobayashi T, Iino M. Transformation of benign fibrous histiocytoma into malignant fibrous histiocytoma in the mandible: Case report. J Oral Maxillofac Surg 2011;69:e285-90.