

Benign fibrous histiocytoma: A rare case involving jaw bone

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Abstract

Benign fibrous histiocytoma (BFH) is a soft tissue neoplasm which occurs mostly on the skin of extremities. BFH rarely occurs in bone and may affect femur, tibia, and pelvic bone. Jaw bone involvement is very unusual with only 11 cases reported till date. This report describes a case of BFH occurring in a 30-year-old female patient affecting left mandibular posterior region. Computed tomography revealed a well-defined expansile lytic lesion in the posterior mandible. Gross examination of the tumor revealed an admixture of fibroblasts and histiocytes in a fascicular and storiform pattern. Immunohistochemical staining was positive for CD68.

Keywords: Benign fibrous histiocytoma, benign tumors, malignancy

Introduction

Benign fibrous histiocytoma (BFH) is a soft tissue tumor originating from mesenchyme which occurs primarily on the skin of extremities.^[1] The etiology of this entity is still obscure. BFH occurs rarely in bone and most of the cases were reported in the femur, tibia, and pelvic bone. Jawbone involvement is even rare. We report one such case of BFH occurring in left mandibular region in a 30-year-old female patient which was asymptomatic since 1-year.

Case Report

A 30-year-old female reported to Department of Oral Medicine and Radiology with a chief complaint of swelling in left lower back tooth region since 1-year. History revealed that patient had undergone extraction of mobile teeth in the same region, 1½ years back. After 5–6 months of extraction, she noticed a swelling in the same region. The swelling progressively increased in size and was associated with mild intermittent pain in left ear. There was no history of pain or


any pus discharge in the associated area. Her past medical and family history was noncontributory. Extra-orally, mild facial asymmetry was noticed on the left side of face near the angle and ramus of the mandible. On palpation, a hard nontender swelling measuring approximately 2 cm × 2 cm was evident on left lower third of face extending from mid body of the mandible to 1 cm anterior to angle of the mandible. On intraoral examination, lower left molars were missing. Mucosa over the alveolar ridge was normal in color and texture. On palpation, 3 cm × 2 cm hard, nontender expansion of buccal and lingual cortices was evident extending from region distal to lower left second premolar up to the anterior border of ramus of mandible with obliteration of the lower buccal sulcus [Figure 1].

Panoramic radiograph presented a multilocular radiolucency involving the left side of the mandible [Figure 2]. Computed tomography (CT) revealed a well-defined expansile lytic lesion involving left angle and ramus of mandible with associated thinning of the cortices and breach in the lingual cortex of left ramus of the mandible [Figures 3 and 4]. Contrast magnetic resonance imaging (MRI) showed that lesion was heterogeneously hyperintense on T2-weighted images, isointense on T1-weighted images with heterogeneous enhancement on contrast. Minimal soft tissue extension was noted along the floor of the mouth abutting the left mylohyoid muscle. The left inferior alveolar nerve (IAN) in the mandibular canal was also seen to be displaced by the lesion [Figure 5].

Based on the painless and slow growing nature of the swelling, located in posterior mandible and existing radiographic appearance, the following conditions were

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Figure 1: Expansion of buccal and lingual cortices in 37–38 regions

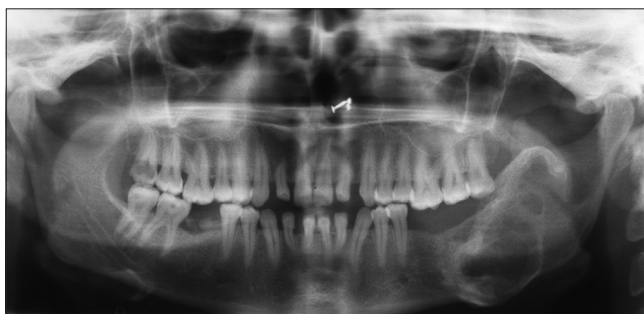


Figure 2: Panoramic radiograph showing well-defined mixed radiolucent radiopaque lesion

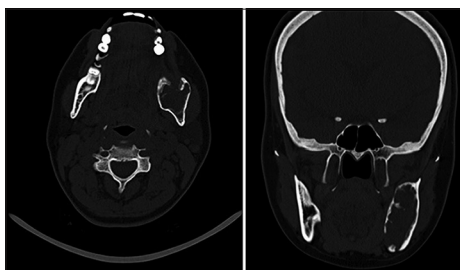


Figure 3: Computed tomography imaging: A well-defined expansile lytic lesion involving left angle and ramus of mandible with associated thinning of the cortices and breach in the lingual cortex of left ramus of the mandible



Figure 4: Computed tomography imaging: Three-dimensional reconstruction

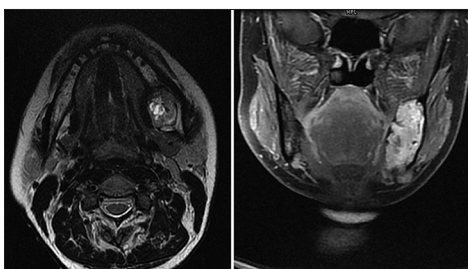


Figure 5: Contrast magnetic resonance imaging showed that lesion was heterogeneously hyperintense on T2-weighted images, with heterogeneous enhancement on contrast

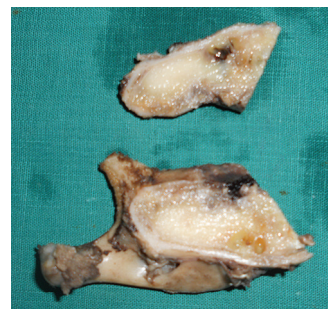


Figure 6: Gross specimen of tumor showing well-defined tumor mass circumscribed within the bone

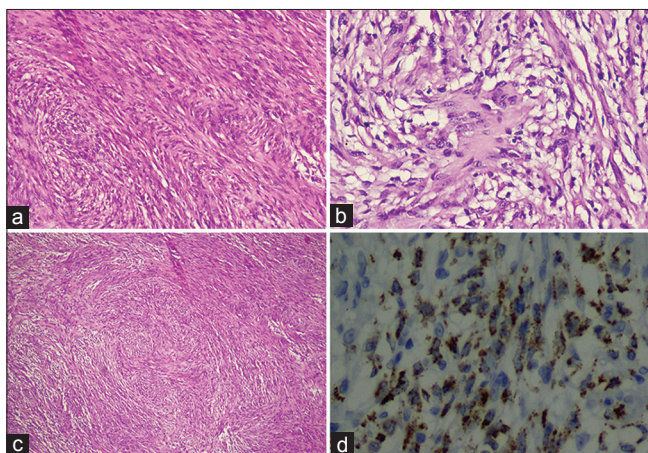


Figure 7: (a) Presence of foamy histiocytes (H and E, $\times 10$); (b) foamy histiocytes and giant cells (H and E, $\times 20$); (c) spindle-shaped fibroblasts and histiocytes seen in storiform and fascicular pattern (H and E, $\times 4$); (d) CD68 positive cells ($\times 40$)

considered in our differential diagnosis such as benign tumors of odontogenic origin such as ameloblastoma, ameloblastic fibroma, odontogenic myxoma, and keratocystic odontogenic tumor. Nonodontogenic tumors such as ossifying fibroma, osteoblastoma, desmoplastic fibroma, intraosseous mucoepidermoid carcinoma, and BFH were considered because of the predilection for occurrence in the mandible with a similar appearance.

The patient was operated under general anesthesia, and segmental mandibulectomy was performed using intraoral approach. Gross examination of the tumor revealed well encapsulated tumor mass with areas of firm but unmineralized yellow tan tissue and partially hemorrhagic red brown tissue [Figure 6]. The microscopic examination revealed an admixture of spindle-shaped cells (fibroblasts)

and epithelioid cells (histiocytes) in a fascicular and storiform pattern. Multinucleated cells and scattered lymphocytes were also seen. The stroma consisted of large areas of foamy cells interspersed with fibrovascular septations. No mitotic activity, cellular atypia or pleomorphism of tumor cells was evident. Vascularity of tumor varied from relatively inconspicuous vessels to exceedingly prominent vascularity with striking hyalinization [Figure 7a-c]. The histopathological features were suggestive of BFH. Immunohistochemical staining showed positive CD68 cells in the lesional tissue [Figure 7d].

The patient has been followed up periodically, but no recurrence or other changes were noted for 2 years.

Discussion

BFH affecting the oral and maxillofacial region is very rare. To the best of our knowledge, only three cases of BFH affecting maxilla and eight cases affecting mandible have been reported in the literature. Among the eight mandibular BFH, five involved posterior region,^[2-6] two involved condylar region^[7,8] and one extended from mandibular right incisor to the left premolar region.^[9] In all the cases, age ranged from 30 to 50 years, with no significant gender predilection. The most common complaints were pain or a long-standing swelling indicating a slow growing nature of this entity. Our patient reported of mild ear pain which may be due to pressure on the auriculotemporal nerve, branch of IAN.

Buccolingual expansion with multilocular radiolucency and a sclerotic rim around the osteolytic defect were typical features in cases reported in the posterior region of the mandible. A similar kind of appearance was evident in our case. There are very few case reports explaining the CT and MRI features of this entity. In CT scan, an area of bony destruction with sclerosis at periphery and breach in the lingual cortex was evident in our case. MRI depicted heterogeneously hyperintense mass on T2-weighted images, isointense on T1-weighted images with heterogeneous enhancement on contrast.

Histological characteristics of BFH in bones are similar to those of BFH occurring in soft tissues. Proliferating fibrohistiocytic cells arranged in a storiform pattern interspersed with foam cells and giant cells is the hallmark of this tumor. Histological findings of BFH and nonossifying fibroma (NOF) are generally overlapping, so clinical and radiographic features may be helpful to differentiate between the two. NOF is seen during the early growth periods and it usually affects the metaphysis of long bones. On the contrary, BFH has been reported in middle age individuals in nonlong bones. Immunohistochemistry can be used to identify the presence of CD68 which is found in the cytoplasmic granules of a range of different blood cells and myocytes. It is particularly useful as a marker for the various cells of the macrophage lineage including monocytes, histiocytes, giant cells, Kupffer cells, and osteoclasts. Recently, KP-1 (CD68) has

been described as a monoclonal antibody to a cytoplasmic epitope present on tissue histiocytes and macrophages.^[10]

BFHs are generally locally aggressive and tend to recur after curettage, hence phenol or liquid nitrogen should be applied to bony walls before bone grafting to clear the residual microscopic defect. Local excision followed by bone grafting is the treatment of choice. Wagner *et al.* had used piezoelectric-assisted cutting device for removal of BFH involving mandible to prevent dentoalveolar nerve injuries in critical cases.^[6] Recurrence of this entity has been reported in several cases with a mean period of 1–2 years after treatment.

Conclusion

BFH of the jaw bone is an exceptionally rare entity with only 11 cases reported till date; the exact nature of the lesion is still not certain. Clinically and radiographically, it mimics the commonly occurring benign tumors of the jaw bone and should be considered in differential diagnosis. Small biopsy samples may be misleading in some cases. A long-term follow-up is required because of its local aggressive nature, high frequency of recurrence and likelihood of transformation to malignant form.

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Conflicts of interest

There are no conflicts of interest.

References

1. Stout AP, Lattes R. Tumors of the soft tissues. Atlas of Tumor Pathology. Second series. Washington DC: United States Armed Forces Institute of Pathology; 1967. p. 38-52.
2. Remagen W, Nidecker A, Prein J. Case report 359: Gigantic benign fibrous histiocytoma (nonossifying fibroma). *Skeletal Radiol* 1986;15:251-3.
3. White RD, Makar J Jr. Xanthofibroma of the mandible. *J Oral Maxillofac Surg* 1986;44:1010-4.
4. 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.
5. Ou DM, Zheng GS, Liao GQ, Su YX, Liu HC, Liang YJ. Clinical and pathologic characteristics and surgical management of benign fibrous histiocytoma of the mandible: A case report. *J Oral Maxillofac Surg* 2012;70:2719-23.
6. Wagner ME, Rana M, Traenkenschuh W, Kokemueller H, Eckardt AM, Gellrich NC. Piezoelectric-assisted removal of a benign fibrous histiocytoma of the mandible: An innovative technique for prevention of dentoalveolar nerve injury. *Head Face Med* 2011;7:20.
7. Heo MS, Cho HJ, Kwon KJ, Lee SS, Choi SC. Benign fibrous histiocytoma in the mandible. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2004;97:276-80.
8. Katagiri W, Nakazawa M, Kishino M. Benign fibrous histiocytoma in the condylar process of the mandible: Case report. *Br J Oral Maxillofac Surg* 2008;46:e1-2.
9. Ertas U, Büyükkurt MC, Çiçek Y. Benign fibrous histiocytoma: Report of case. *J Contemp Dent Pract* 2003;4:74-9.
10. Binder SW, Said JW, Shintaku IP, Pinkus GS. A histiocyte-specific marker in the diagnosis of malignant fibrous histiocytoma. Use of monoclonal antibody KP-1 (CD68) *Am J Clin Pathol* 1992;97:759-63.