CASE REPORT

Occult massive ossifying fibroma of maxilla extending into the maxillary antrum – A rare case report

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Abstract

Aim: The aim of this study was to report a peculiar case of massive ossifying fibroma which had an occult presentation, occupying the maxillary antrum. Background: Ossifying fibroma is a slow-growing benign fibro-osseous lesion that most commonly affects the mandible (70–80%). When it involves the maxilla, it is more extensive. It may affect the nasal cavity, and thereby causing nasal blockage and breathing difficulty even without extensive asymmetry. Case Description: The patient complained of swelling in the left upper posterior teeth region for the past 6 months. A history of nasal blockade and difficulty in breathing was present. Deviation of tip of the nose was evident without gross facial asymmetry. Intraorally, the swelling extended buccopalatally, crossing the midline with smooth surface. Radiological investigation revealed mixed radiolucency and radiopacity with distinct borders with apical root resorption of involved teeth and bicortical expansion was evident. The extent of the lesion involved the entire left maxillary antrum, airway of the nasal cavity, and pushing the nasal septum to the contralateral side. Surgical enucleated was performed. Conclusion: Six months postoperatively, the patient showed uneventful healing with evidence of bone formation with relief from symptoms. Surgical enucleation has demonstrated a good prognosis in this case, as ossifying fibromas are encapsulated and well demarcated from surrounding tissues. Clinical Significance: This case report documents a peculiar case of extensive ossifying fibroma of the maxilla, involving the sinus that hinders breathing and showing minimal asymmetry. Prompt diagnosis and treatment have proven effective with no complications postoperatively.

Keywords: Bone neoplasm, fibro-osseous lesion, maxilla, ossifying fibroma, sinonasal

Introduction

Fibro osseous lesions are a group of entities among face and jaw disorders. There is a replacement of bone by a benign connective tissue matrix and varying mineralized substances. Types being cemento-osseous dysplasia, fibrous dysplasia, and ossifying fibroma. The World Health Organization, in 1992,[1] classified an ossifying fibroma as a “demarcated or rarely encapsulated neoplasm consisting of fibrous tissue containing varying amounts of mineralized material resembling bone and/or cementum.” When the lesion contains cementum, it is called as cementifying fibroma, in the case of bone, called as ossifying fibroma; if both are present, called as cemento-ossifying fibroma.[2] It occurs between the third and fourth decades of life with a propensity among females (4:1), prevailing in the mandibular premolar-molar region. They are usually slow-growing intrabony mass that is delimited and asymptomatic, though over time, they may become large enough causing facial deformation.[3] Small lesions seldom cause any symptoms. Painless swelling of the bone involved and obvious facial asymmetry is seen in larger lesions. Some of the lesions behave aggressively, even reach massive proportions with voluminous cortical expansion. The aggressiveness of the lesion is more common in children and has led to the designation of juvenile aggressive OF or active ossifying fibroma.[4]

Displacement and resorption of tooth roots are observed sometimes. The ossifying fibroma is not commonly associated with pain and paraesthesia. Early lesions are small and radiolucent. However, as the lesions mature, they become mixed radiolucent and radiopaque and then transforms ultimately into the radiopaque lesion.[5] Downward bowing of the inferior cortex of the mandible is often characteristically seen in large lesions. Histopathologically, it comprises fibrous tissue with an inconstant degree of cellularity and contains mineralized material in the form of trabeculae of osteoid and bone or basophilic and poorly cellular spherules, resembling cementum. The admixtures of the two types are typical. The bony trabeculae...
that are present vary in size and often demonstrates a mixture of lamellar and woven patterns. Here, we are reporting a peculiar case of extensive ossifying fibroma involving the maxilla and sinus, hindering breathing, and showing minimal asymmetry.

**Case Report**

A 35-year-old male patient reported to our institution with a chief complaint of swelling in the left upper posterior teeth region for the past 6 months. The patient was asymptomatic 6 months, before 1 day, he noticed swelling in the left upper back tooth area which was gradual in onset with size the same as it was noticed and was not associated with pain and paraesthesia. The patient has history of the left nostril blockage and difficulty in breathing for 10 months with a change in voice. An extraoral examination revealed no gross facial asymmetry. There was deviation of tip of nose to right side, and tenderness elicited on left maxillary sinus area. Bilateral submandibular lymph nodes enlarged, one on each side, approximately 0.5 × 0.5 cm, soft to firm in consistency, non-tender, and mobile. Intraorally, a single diffuse swelling was presented extending buccally over left maxillary premolars and molars, with obliteration of buccal vestibule extending from teeth 25 till 28. On the palatal aspect, the swelling was extending from the distal aspect of the left maxillary canine till distal aspect of the third molar, crossing of midline and mucosa appearing normal to adjacent mucosa [Figure 1]. Hard tissue examination revealed mobile left maxillary molars and tenderness on percussion of the second premolar and first molar. Routine laboratory investigations were done and unremarkable.

Orthopantamogram [Figure 2] revealed mixed radiolucent radiopacity present extending from the left maxillary central premolar till distal aspect of the third molar. The haziness of the left maxillary sinus was evident when compared to the contralateral side. The inferior and medial wall of the sinus cannot be traced. Paranasal sinus view [Figure 2] revealed homogenous radiopacity in the left maxillary sinus when compared to contralateral side. Medial, inferior, and lateral wall of sinus cannot be traced and superior border was intact.

Cone-beam computed tomography (CBCT) axial section [Figure 2] revealed a mixed hyperdense hypodense area in the left maxilla extending from periapex of lateral incisor till the third molar, with the buccal and palatal cortical expansion with thinning. CBCT superior axial section revealed the narrowing of nasal fossa and maxillary sinus space. CBCT sagittal section further revealed apical root resorption in premolars and molars.

CT axial section bone window [Figure 2] revealed a mixed hyperdense hypodense region in the left maxilla extending from 21 till 28 with the bicortical expansion with thinning of the cortex, involving the midline and extending into the left nasal fossa, deviating nasal septum, and expands extensively into the maxillary sinus, measuring 49 mm × 41 mm in its greatest dimension.

Incisional biopsy was done under local anesthesia, and histopathology report revealed “ossifying fibroma.” Based on the findings, surgical removal of fibroma was opted under general anesthesia. By the intraoral approach, a triangular flap was raised with vertical releasing incision ir 22. The extraction of canine, premolars, and molars was done. Lesion was relieved from all attachments and enucleated as a total mass [Figure 3]. Thorough irrigation was done with betadine and saline. Gel foam placed and closure done with 3-0 vicryl. The specimen was sent to
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Histopathological study and was confirmed to be “OSSIFYING FIBROMA.” The soft tissue was predominantly fibrous in nature with some areas showing plumpy fibroblasts which were relatively less vascular in nature. Within fibrous tissue, there were round calcified structures noticed, which are homogenous in nature giving eosinophilic appearance [Figure 4]. There is no evidence of mitosis or abnormality of fibroblastic cells. The patient has been kept under follow-up and 6 months, no clinical signs and symptoms and post-operative CBCT revealed bone formation of about 6.2–7 mm linear dimension in the surgical site [Figure 5]. The patient kept under observation for further prosthetic rehabilitation.

Discussion

Benign fibro-osseous neoplasms are described by their constituents, such as ossifying fibroma, in which bone outweighs, while the term cementifying fibroma is used when curved/linear trabeculae or spheroidal (psammoma-like) calcifications are detected. If in case both bone and cementum-like material are noticed, then the lesion is assigned as cemento-ossifying fibromas.[1]

Management of fibro-osseous lesions should not be generalized and must be done by assessing the clinical evolution of each case and further considering the benign nature and growth pattern.[5] The treatment options are grouped as conservative enucleation, curettage, and radical surgery.

On the basis of an analysis of 64 cases classified as ossifying and/or cementifying fibroma, Eversole et al.[5] concluded that a distinction between these two variants would be academic, as no behavioral or histological differences exist. The tumor is relatively hypovascular and well-demarcated from the surrounding tissue, which permits comparably easy separation from the surrounding bone during surgical intervention. Some lesions will have a definite capsule. This demarcation from the surrounding tissue is an important feature in distinguishing ossifying fibroma and fibrous dysplasia.[6] Surgical removal and curettage are the preferred treatment of choice since the tumor is well-delimited. In the case of very large lesions with important tissue ablation, the challenge is to replace the affected tissue.[6] The circumscribed nature of the lesion permits the local enucleation or curettage of smaller lesions.[5] The treatment of ossifying fibroma is excision of the whole lesion with a rim of surrounding normal tissues, as suggested in the literature. However, management should be individualized for each case, depending on the size, location, benign nature, and growth behavior of the lesion.

In the present case, we selected conservative enucleation of the lesion with thorough debridement; curettage of the bone bed was done to promote the good healing of the defect. A study by MacDonald-Jankowski[8] supports the enucleation or curettage as the first treatment option, and it affords a recurrence rate of 0–28% if relapse is identified in the course of follow-up, conservative resection is obligate. Ossifying fibromas have a predominantly good prognosis, with less recurrence rate. However, data regarding the prognosis of sinonasal ossifying fibroma remain elusive. Aggressive ossifying fibromas show a recurrence rate of about 30–38%. In this case, even though benign lesion, the site, occult extension, and hindrance in olfactory functions made it peculiar. Prompt treatment has efficiently prevented further involvement of the nasal fossa and maxillary sinus, reversing patient’s breathing difficulties.

Conclusion

Even though ossifying fibromas seem to be benign and slow-growing, some of them may show occult presentation, especially...
in the maxilla. Legitimate diagnosis and surgical enucleation of the lesion have resulted in the success of the management and relieved the patient from the symptoms.

**Clinical Significance**

In this case, the patient noticed difficulty in breathing and stuffiness, even before the occurrence of noticeable swelling. Relative signs should be remarked, and the risk must be assessed. Surgical enucleation of ossifying fibroma implies to be effective with no post-operative eventualities and recurrence since they are encapsulated and well-demarcated from adjacent tissues.

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