OSSIFYING FIBROMA OF THE MAXILLA: CASE REPORT.

Dr. Kalaiselvi Santhosh [*], Dr Anu Sushanth. A [**]

Abstract

Fibrous dysplasia, ossifying fibroma (OF),cemento-ossifying fibroma, florid osseous dysplasia, and localised osseous dysplasia are all fibro-osseous lesions in the jaw bones. The most frequent fibro-osseous lesion is OF, which appears as a slow-growing, well-encapsulated benign neoplasm made of various quantities of bone or cement-like tissue in a fibrous stroma well-demarcated from the neighbouring normal bone. OF is commonly found in the jaw bones, with a preference for the mandible. OF in a patient usually appears as a single lesion and less occasionally as numerous lesions. We discuss clinical features and radiologic findings, histology, and surgical care of a rare case maxilla, as well as a brief literature review.

***Key words**: Fibrous dysplasia, ossifying fibroma (OF), Cemento-ossifying fibroma, Florid osseous dysplasia*

Fibrous dysplasia, ossifying fibroma (OF), florid osseous dysplasia, cemento-ossifying fibroma (COF), and localised osseous dysplasia are all examples of fibro-osseous lesions (FOL) of the jaw bones.[1]The most common fibro-osseous lesion is OF, which appears as a slow-growing, encapsulated benign neoplasm composed of various quantities of bone or cementum-like tissue in a fibrous stroma, well-demarcated from neighbouring normal bone.[2] It is most frequent in the jaw bones, particularly the mandible. Because OF contains cementum-like tissue and bone, the terms OF or COF are sometimes used to characterize this tumour. However, the consensus is that both names represent the same underlying histology and so describe the same type of lesion.[1]

CASE REPORT

A 53-year old male patient came to dental OP with the chief complaint of a painless swelling in the left mid face region for the past 1 year. History of presenting illness revealed an asymptomatic swelling which started 1 year back on the left side mid-face region. It gradually increased to attain the present size. It is not associated with pain, and/or fever. No history of secondary changes. He had taken medication for about one week,but the swelling didn't subside. On general examination it was revealed that the patient was afebrile with gross facial asymmetry due to a single, smooth surfaced, well-defined swelling, which was roughly oval in shape and approximately 3 × 2 cm in size, seen in the left middle third region of the face. Overlying and surrounding skin was normal. Swelling extends superiorly 2cm below the infraorbital margin, inferiorly 1cm below line joining angle of the mouth to pinna of ear, medially up to lateral extend of nose, obliterating the naso-labial fold. There was no secondary changes. On palpation, swelling was not warm. Inspection findings with respect to size, shape, margin, and surface were confirmed. The swelling was non tender, hard in consistency and not mobile.

Intra-Oral Examination: Single, smooth surfaced, well defined swelling measuring 3×2cm in size, roughly oval in shape presented in the left side palatal region. Swelling was extending anteriorly from mesial aspect of 24, posteriorly to distal aspect of 27; medially till mid line of the palate, laterally till attached gingiva of 24, 26, and 27. Colour appeared pale, surrounding mucosa and mucosa over the swelling were normal. No visible pulsation or secondary changes. On palpation inspection findings with respect size, shape, margin, extent and surface were confirmed. The swelling was non tender, hard in consistency. Bi-cortical expansion was elicited.

Considering an old aged male patient, with a complaint of a long standing asymptomatic swelling on his upper left back teeth region which was hard in consistency, we provisionally diagnosed as a benign lesion.

[*] Reader, Department Of Oral Medicine and Radiology Karpaga Vinayaga Institue Of Dental Sciences , Maduranthakam, Chengalpattu, Tamil Nadu.
[**] Reader, Department Of Oral Medicine and Radiology, Educare Institute of Dental Sciences, Malappuram, Kerala.

Correspondence address: dr.sks13@gmail.com

The differential diagnoses of fibro-osseous lesions were considered. Fibrous dysplasia is more common in the maxilla and it tends to grow longitudinally with ill-defined margins, in contrast to OF, which is predominantly seen in the mandible, usually encapsulated, and has a centrifugal growth pattern

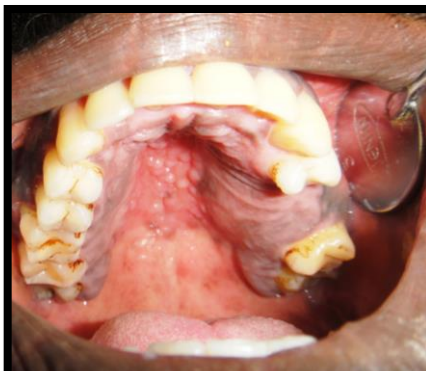

Fig. 1 Swelling of the left palatal region

Upon examination of IOPA, it was observed that there was loss of periodontal ligament space and lamina dura in relation to 25, 26, and 27. OPG showed unilateral abnormality in alveolar bone of left maxillary premolar and molar region and the epicenter lies at the periapical region of 25 to 28. Lesion has a smooth, well-defined outline without a sclerotic border. And internal structures were uniformly radiopaque.

Axial section of CT showed a homogenous hypodense mass extending from distal aspect of 24 to 28 region with average of 85HU. Elective surgical subperiosteal partial en-bloc resection was performed, from 24 to 27.Immediate reconstruction was performed with a microvascular iliac bone flap.

Histopathological examination revealed highly cellular connective tissue stroma showing plump fibroblasts. The fibroblast showed oval nuclei with dispersed chromatin and indistinct cytoplasm. Numerous trabeculae of new bone formation were seen along with limited number of small hematoxyphilic calcifications dispersed throughout the lesions. Periphery showed partial capsulation. Based on the histopathology, diagnosis of “Ossifying Fibroma” was given.

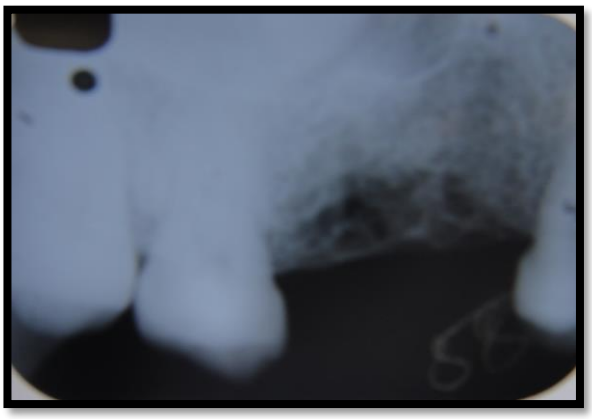

Fig. 2. IOPA showing indistinguishable lamina dura and periodontal ligament space.

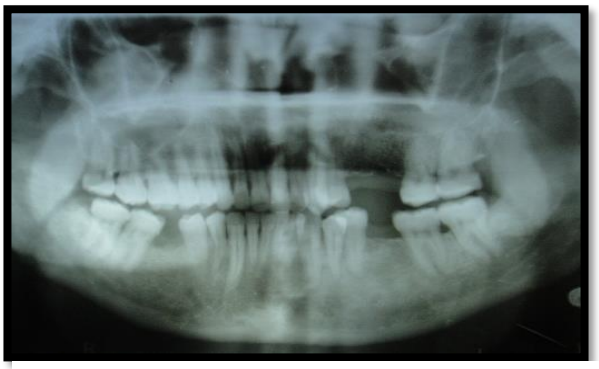

Fig. 3-OPG shows well defined uniform radiopacity in 25 to 28 region

Discussion:

FOL are an uncommon class of lesions with a pathogenesis that is unknown. OF is a benign bone lesion that most frequently affects children and is located in the mandible and maxillary sinus (75-89%). The first instance of OF was reported to Menzel in 1872 as a rare benign primary cranial skeleton bone tumour that most frequently affects the jaws.[1]

Odontogenic tumours were described in the World Health Organization's classification of head and neck tumours, 4th edition and maxillofacial bone tumours, group OF with familial gigantiform cementoma, fibrous dysplasia, cemento-osseous dysplasia, and osteochondroma under fibro and chondro-osseous lesions. [2]

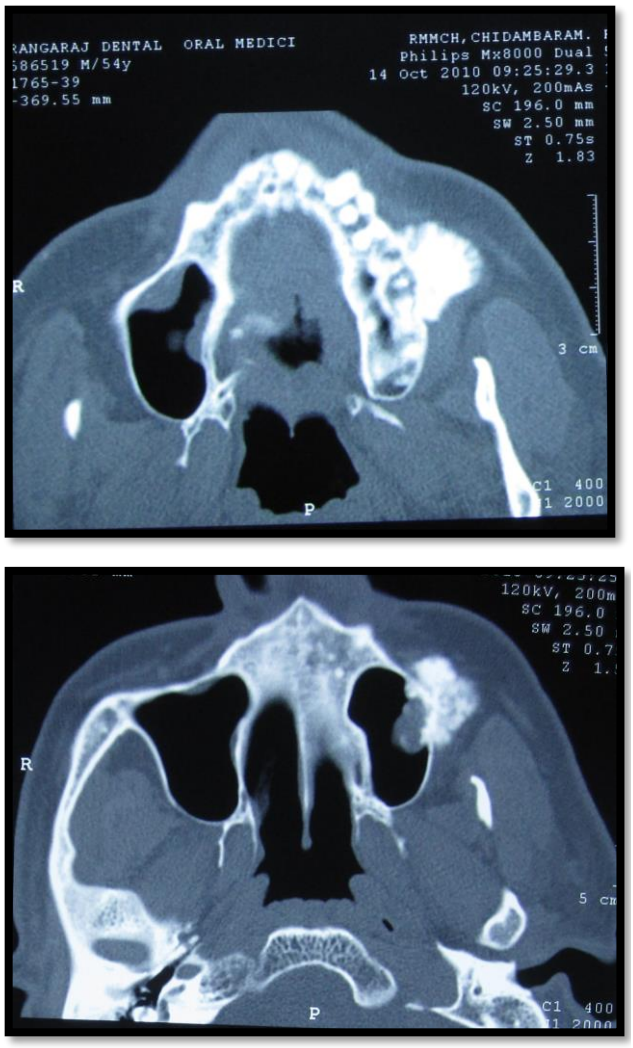

Fig. 4 Axial view- Homogenous hypodense

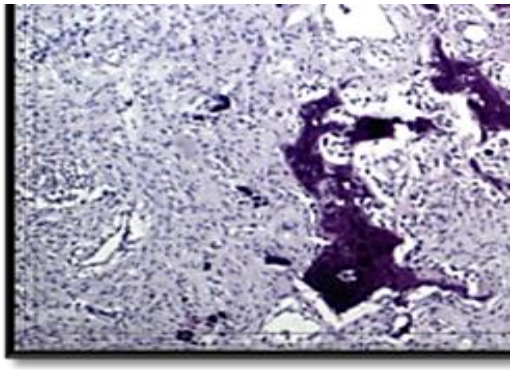

Fig. 5 Trabeculae of new bone formation seen along with limited number of small hematoxyphilic calcifications

.

COF, juvenile trabecular OF (JTOF), and juvenile psmammomatoid OF (JPOF) are the three types of OF that are recognized. The origin of COF is odontogenic, and it is slow-growing with a preference for the posterior mandible and infrequently the maxilla in females.[1] JPOF is uncommon, with a mean age range of 16 to 33 years, and predominates in the orbit and ethmoid bones and the craniofacial bones in the extragnathic regions.[3] JPOF is more aggressive, exhibits a well defined periphery with a mixture of radiopacity and radiolucency. Complete resection is the only method of treatment for JTOF and JPOF, albeit recurrence is more common than in COF. 70% of cases of OF involves the mandible, with the remaining 22% occurring in the maxilla's molar region, the ethmoids, the orbital regions, and in very rare cases the petrous bone.[2]

COF frequently manifests as a single lesion, although it is not uncommon to observe several lesions or cases with multiple, familial, or successive presentations.[4] There are many theories put out on the pathogenesis of OF. Mesenchymal cells with pluripotency, which can grow into cementum, bone, and fibrous tissue, are the source of OF.[2,4] Both an excessive proliferation of periodontal ligament cells and a metastatic process occurring in connective tissue fibres (non-periodontal in origin) have been proposed as potential causes.[5] Because of the considerable mesenchymal cellular induction into bone and cementum during odontogenesis, the jaws are frequently damaged.[6] Periodontitis, past extractions, trauma, and potential genetic abnormalities are all being monitored currently. A predisposition for the mandible and infrequently the maxilla, OF can develop at any age, but it is most common in children and young adults, with a female predominance in the third and fourth decades of life. [7,8]

The definitive diagnosis of OF is aided by magnetic resonance imaging (MRI) and CT scans. Typically, round or oval, relatively smooth, well-defined, expansile mass with corticated border and variable degree of internal radiopacity are the radiographic characteristics of OF.[9] The inner aspect may be radiolucent or have a radiolucent periphery that represents a fibrous capsule,

cortical bone expansion, or a combination of radiolucent and radiopaque tissue. The inner aspect may be granular, as in fibrous dysplasia. The mandibular canal is typically shifted inferiorly around the tumour. A well-defined unilocular mixed lesion with a ground-glass appearance internally and a thin radiopaque edge can be seen on OPG, and root resorption may be present. [10]

With a peripheral radiolucent area in the maxilla and mandible, the OPG in our case revealed extensive, well-encapsulated, well-demarcated lesions that were mixed radiolucent and radiopaque. An expanded affected bone can be seen on a coronal cone-beam computed tomography (CBCT) along with a concentric corticated lesion and radiopaque foci. A well-defined radiolucent lesion can be seen on a sagittal CBCT, and an enlargement of the bone can also be seen on an axial CBCT.[2] In the axial view of a digital volumetric tomography (DVT) scan, undamaged cortical bone may be seen expanding and thinning along with radiopaque calcification within an interior structure.

A 3D Recon scan reveals mixed density caused by varying amounts of radiopaque material. The characteristic CT appearance of OF is an expansile, well-circumscribed lesion that resembles ground glass and may or may not have a cystic component.[11] In our case, axial and coronal CT scan images revealed an expansile osteogenic lesion in the maxilla (left maxillary lesion extending distally to the third molar to the left temporomandibular joint and showing well-delineated spherical margins and a temporomandibular joint lesion) and in the mandible (extending from distal to the third molar to the left orbit and left lateral nasal wall). On precontrast T1-weighted pictures,

MRI displays intensities resembling brain grey matter, and on T2-weighted sequences, intermediate to low levels; after gadolinium injection, there is an amplification of the lesion. [12]

Histologically, OF appears as a moderately avascular fibrous stroma with spheroidal calcifications that resemble cement structures and fusiform cells mixed in with bone trabeculae. [13] There could be multinucleated giant cells. The calcified material is made up of trabeculae of woven bone with irregular shapes, lamellar bone trabeculae that are dispersed, deposits of basophilic staining that are round or oval, cellular or acellular calcified deposits that have been compared to cementum, or any combination of these.[4] Microscopically, OF is composed of cellular connective tissue that is mineralized and has osteoblastic rimming on the surface. This can be compared to our case's histology and microscopy.

Depending on the extent of the lesion, complete surgical removal using curettage, surgical excision, or en-bloc resection is the treatment plan.[12] In order to lower the likelihood of recurrence and the potential for malignant transformation, radical surgery is indicated. The techniques employed were as follows: Enucleation and curettage were performed on 36% (n=09) of the mandible, 32% (n=03) underwent segmental mandibular excision and reconstruction with reconstruction plate, and 20% (n=05) underwent microvascular fibula graft reconstruction. 20% (n=05) of the maxilla underwent enucleation and curettage, whereas 12% (n=03) underwent partial maxillectomy.

Due to the size of the tumour in our situation, three-quarters of the maxilla had to be removed. Recurrence results from incomplete or partial resection.[2] The "recurrence" mentioned in our situation may have been caused by insufficient enucleation. After surgery, the recurrence period can happen at any time and might last anywhere between six months and seven years. As a result, a follow-up term of up to 10 years is advised.

CONCLUSION

Histopathological, clinical-radiological, and other investigations can help make an accurate diagnosis. It is uncommon for the maxilla and mandible to move synchronously at different places. While surgical excision is sufficient in the majority of cases of OF, radical surgical resection is recommended for extensive lesions. Extensive en-bloc resection is recommended for lesions that are large in order to lower the likelihood of recurrence.

References

1. Bhat P, Kaushik A, Vinod VC, Sridevi K, Chaudhry A: Ossifying fibroma of the maxilla: a rare case . Int J Radiol Radiat Ther. 2017, 3:198-201. 10.15406/ijrrt.2017.03.00054
2. Chidzonga M, Sunhwa E, Makunike-Mutasa R (January 27, 2023) Ossifying Fibroma in the Maxilla and Mandible: A Case Report With a Brief Literature Review. Cureus 15(1): e34257. DOI 10.7759/cureus.34257
3. Makkad RS, Naidu GS, Nagi R, Sagtani A, Patil S, Shrivastava S: Multiple fibro-osseous lesions of the jaws: areport of a rare case with a literature review. Imaging Sci Dent. 2021, 51:461-6. 10.5624/isd.20210021
4. Rabelo N, da Silva VT, do Espírito Santo MP, et al.: Orbit ossifying fibroma - case report and literature review. Surg Neurol Int. 2020, 11:35. 10.25259/SNI_492_2019
5. Liu Y, Wang H, You M, Yang Z, Miao J, Shimizutani K, Koseki T: Ossifying fibromas of the jaw bone: 20 cases. Dentomaxillofac Radiol. 2010, 39:57-63. 10.1259/dmfr/96330046
6. Wang TT, Zhang R, Wang L, Chen Y, Dong Q, Li TJ: Two cases of multiple ossifying fibromas in the jaws . Diagn Pathol. 2014, 9:75. 10.1186/1746-1596-9-75
7. Pick E, Schäfer T, Al-Haj Husain A, Rupp NJ, Hingsammer L, Valdec S: Clinical, radiological, and pathological diagnosis of fibro-osseous lesions of the oral and maxillofacial region: a retrospective study. Diagnostics (Basel). 2022, 12:238-44. 10.3390/diagnostics12020238
8. Jih MK, Kim JS: Three types of ossifying fibroma: a report of 4 cases with an analysis of CBCT features . Imaging Sci Dent. 2020, 50:65-71. 10.5624/isd.2020.50.1.65
9. Bhat SV, Kumar SP, Periasamy S, Krishna VK: An uncommon presentation of ossifying fibroma in the maxilla. Cureus. 2022, 14:e23638. 10.7759/cureus.23638
10. Rahman MA, Rahman T, Halder IA: Clinicopathological study of ossifying fibroma . Update Dental College Journal.2019. 9:7-13. 10.3329/updcj.v9i2.43732
11. Hwang EH, Kim HW, Kim KD, Lee SR: Multiple cemento-ossifying fibroma: report of an 18-year follow-up . Dentomaxillofac Radiol. 2001, 30:230-4. 10.1038/sj.dmfr.4600608
12. Bertolini F, Caradonna L, Bianchi B, Sesenna E: Multiple ossifying fibromas of the jaws: a case report . J Oral Maxillofac Surg. 2002, 60:225-9. 10.1053/joms.2002.29832
13. Canger EM, Celenk P, Kayipmaz S, Alkant A, Gunhan O: Familial ossifying fibromas: report of two cases . J Oral Sci. 2004, 46:61-4. 10.2334/josnusd.46.61

Conflict of Interest: *None Declared*