

Focal Cemento-osseous Dysplasia: Report of an Unusual Case

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Abstract:

Fibro-osseous lesions of the jaws pose diagnostic and therapeutic difficulties due to their clinical, radiological and histological variability. Cemento-ossifying fibroma is a rare, benign fibro-osseous lesion that arises from the periodontal ligament. Cemento-osseous dysplasia is another variant of fibro-osseous lesion which commonly involves mandible of females. We report a case of an unusual presentation of cemento-osseous dysplasia in a 13-year-old female which posed a diagnostic dilemma clinically and histopathologically, due to its close resemblance to cemento-ossifying fibroma.

Keywords: Cemento-osseous dysplasia, Cemento-ossifying fibroma, Fibro-osseous lesions

Introduction :

Focal cemento-osseous dysplasia (FCOD) and cemento-ossifying fibroma (COF) are two clinically recognized entities that are not easily differentiated histopathologically because of lack of recognition of specific microscopic features.^{1,2} Both of these fall under the category of fibro-osseous lesions that are characterized by the replacement of normal bone by connective tissue, with varying degrees of mineralization in the form of woven bone or cementum-like round acellular basophilic structures. Both radiologically and histopathologically it is difficult to distinguish between these two entities. However, differentiation of these two lesions is critical because the treatment protocols are quite different. We present case of a 13-year old female patient which posed a diagnostic challenge; as the lesion clinically appeared more as an ossifying fibroma, but the histopathological picture was consistent with cemento-osseous dysplasia.

Case report :

A 13-year-female patient along with her parents, reported with the complaint of swelling on the left side of the face since one month. Swelling was mildly painful and had gradually increased to the present size. Clinical evaluation revealed a large extra-oral, ovoid, mildly tender swelling involving entire left mid and lower facial region.(Fig.1) Borders were diffused, and consistency was bony hard superiorly and firm in the inferior aspect. Orbital floor on the same side also seemed to be raised. Intra-orally, expansion of buccal and palatal cortical plates associated with maxillary left premolar - molar and adjacent distal alveolar region was seen. Obliteration of buccal vestibule was present in the same

region. Another notable finding was the missing maxillary left second molar. (Fig. 2)

Conventional radiographic investigation, which included vater's view and PA skull, revealed diffuse radiopacity obliterating the left maxillary sinus. Displacement of two teeth: one adjacent to the roof of maxillary sinus and second to the infra-orbital region was also seen. (Fig. 3 (a) & (b)) Computed tomography revealed a single, large expansile lesion arising in the left alveolar region, encroaching on the left maxillary sinus. (Fig.4 (a),(b),(c)) Lesion showed amorphous calcification within it and contained two teeth which appeared to be molars. The alveolus showed erosion of both inner and outer cortical plates. Hypodense non-enhancing areas suggestive of secretions were also seen in the lesion. The left orbital floor appeared slightly superiorly displaced, more so posteriorly. (Fig. 5 (a) & (b))

Based on the history, clinical and radiographic features, a differential diagnosis of secondarily infected juvenile ossifying fibroma, focal cemento-osseous dysplasia and adenomatoid odontogenic tumor was thought of.

Incisional biopsy was then carried in which friable, granular and gritty tissue was obtained. (Fig. 6) Histopathological examination revealed a fibrocellular stroma with numerous spindle-shaped fibroblasts arranged in a whorled pattern. Numerous osteoid and cementoid globules with woven bony trabeculae were noted. The extravasated blood elements were also seen in abundance. (Fig. 7 (a) & (b)) Based on the histopathological presentation, a diagnosis of cemento-osseous dysplasia (COD) was made. Though this picture also resembles ossifying fibroma, few distinguishing features were noticed. The trabeculae in ossifying fibroma tend to be more delicate than seen here. The cementum-like particles in ossifying fibroma are more ovoid and often demonstrate brush borders in intimate association with the adjacent

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Fig. 1: Large extra-oral swelling seen on the left side of the face.

Fig. 2: Intra-oral view showing prominent palatal cortical expansion

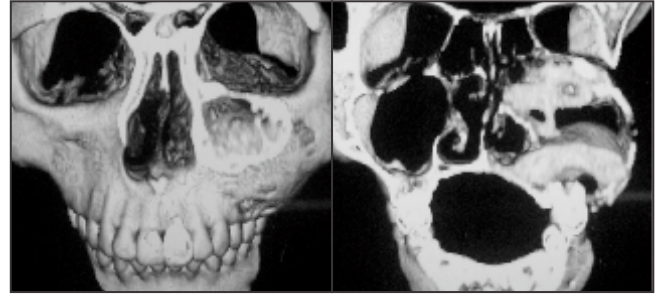


Fig. 5 (a) & (b): 3-D reconstructed CT images

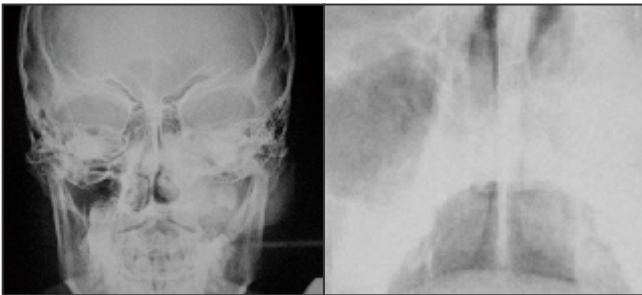


Fig. 3 (a) & (b): PA skull and Water's view showing diffuse radiopacity obliterating the left maxillary sinus. Note the displacement of molars, one adjacent to the roof of maxillary sinus and second to the infra-orbital region



Fig. 6: Intra-operative view during incisional biopsy. Note the prominent buccal cortical expansion

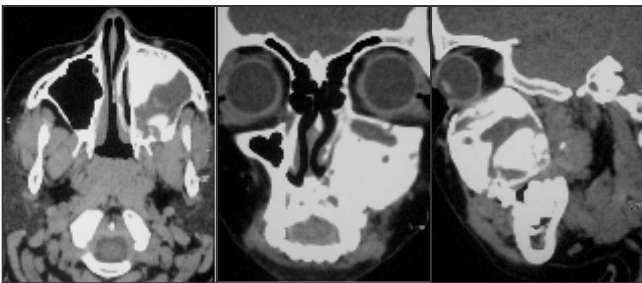


Fig. 4: Computed Tomography images (a) axial, (b) coronal and (c) sagittal view showing a large, expansile lesion with amorphous calcification within it

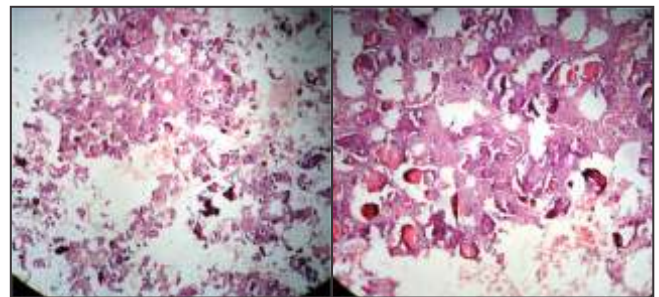


Fig. 7 (a) & (b): H & E staining (4X & 10X) showing histopathological features suggestive of Cemento-osseous dysplasia

stroma. Osteoblastic rimming of trabeculae, typically seen in ossifying fibroma was also not seen in this case. However, extravasated blood elements, typically seen in COD, were present throughout the lesion. Juvenile ossifying fibroma was ruled out, considering absence of any aggressive features such as mitotic figures and cellular hyperactivity. Eventually, a final diagnosis of focal variant of cemento-osseous dysplasia was made.

Patient was then admitted to Bharati Hospital and Research Centre, Pune for surgical management of the lesion. Last contacted, left maxillary resection; followed by grafting procedure was being planned for the patient.

Discussion :

The concept of focal fibrous cemento-osseous dysplasia exhibit was not clarified until the mid-1990s. Before that time, most cases were misdiagnosed as variant of ossifying fibroma. About 90% of cases of FCOD occur in females, with a predilection of third to sixth decades. It may occur in any area of the jaw, but the posterior mandible is the most predominant site. The lesion is largely asymptomatic and rarely becomes larger than 1.5 cm in size. Radiographically the lesion varies from completely radiolucent to densely radiopaque with a thin radiolucent rim.³

The cemento-ossifying fibroma is considered a rare, benign fibro-osseous lesion and true neoplasm with a significant growth potential.^{4,5} Ossifying fibromas occur in a wide age range, with majority of cases seen in third and fourth decades

of life. Premolar and molar regions of the mandible are involved more often than the maxilla. The lesion reveals a painless course, very often only discovered during routine radiologic examination.

The features of FCOD are less specific and often mandate surgical investigation.⁶ Even upon histopathologic review, distinguishing FCOD from ossifying fibroma can be difficult. Su et al in 1997 assessed 20 pathologic parameters for their ability to distinguish reliably between the two entities.⁷ They concluded that majority of FCOD were composed of multiple fragments of granular tissue. "Ginger root" pattern and irregularly shaped cementum like masses were typically seen in FCOD, whereas thin isolated trabeculae with prominent osteoblastic were more commonly seen in COF. Free hemorrhage was frequently interspersed in the artifactual spaces throughout FCOD. These prominent features allowed distinction histopathologically in 94% of cases.

Treatment of FCOD has to be individualized for the patient. For the asymptomatic patients with small lesions, the best management consists of regular recall examination with prophylaxis and good home hygiene. In cases where the lesion is large enough to cause any deformity or discomfort to the patient, radical resection, local excision conservatively or enucleation with curettage may be considered among the treatment alternatives.³

The case reported here had a rather unusual presentation, considering the site, size as well as the histopathological presentation. It also emphasizes the need to develop a consensus on proper diagnostic criteria for lesions like FCOD and COF to avoid any confusion regarding the identification and management.

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