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Cone-Beam CT Evaluation of Mandibular Bilateral Florid Cemento Osseous Dysplasia: Case Report and Literature Review of 5 Years

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Abstract

Fibro-osseous lesions (FOLs) are a category of conditions in which normal bone is replaced by fibrous connective tissue and pathological bone growth occurs. Cemento-osseous dysplasias (CODs) are a group of non-neoplastic lesions that affect the jaws. The categorization of fibro-osseous lesions has long been a

source of contention, and the WHO classification has been revised numerous times. Florid Cemento-osseous dysplasias (FCODs) can be radiolucent (osteolytic phase), mixed (cementoblast phase), or radiopaque (osteogenic phase) and have a narrow radiolucent peripheral halo involving many quadrants. To prevent an excessively protracted clinical course, a comprehensive clinical, radiological, pathological, and biochemical evaluation is required for the identification of such lesions. As a result, in order to better comprehend these circumstances, we present this case report together with a review of the literature from the preceding five years.

Keywords

Classification, Florid cemento-osseous dysplasia, Cone-Beam Computed Tomography, Fibro-osseous lesions, non-neoplastic lesion, bone pathology, mixed radiolucent-radiopaque lesion, Developmental bone diseases, Bone dysplasia, Differential diagnosis

Introduction

Fibro-osseous lesions (FOLs) are a diverse group of lesions with characteristic replacement of normal bone by fibrous connective tissue along with osseous components [1]. Cemento-osseous dysplasias (CODs) are comprised of a spectrum of non-neoplastic FOLs which exclusively occur in tooth-bearing areas of the jaw and based on the extent, classified as focal, florid and periapical variants [2].

FCOD occurs sporadically or as inherited familial variant [2]. Since the lesion is asymptomatic in the absence of infection, it is of utmost importance for clinicians to be aware of this rare entity to prevent misdiagnosis and inappropriate clinical intervention that could lead to a protracted clinical course.

Case Report

A 72 years old female reported with a chief complaint of missing teeth and wanted prosthesis for the same (Figure 1). The past dental history indicated teeth extraction 20 years back in the mandibular anterior region due to trauma. Following the clinical assessment, implant was determined to be the best therapeutic option.

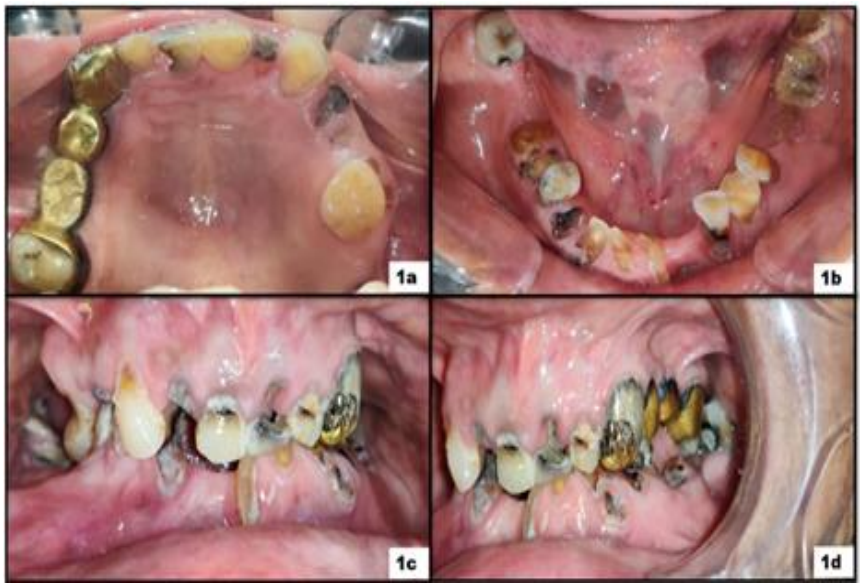

Figure 1: Clinical photographs of the patient with no obvious pathological abnormalities

CBCT examination revealed a massive, expansile, osteolytic lesion spreading from the 48 area to the 42 region, and then more posteriorly from the 32 region to the 38 region (Figure 2).

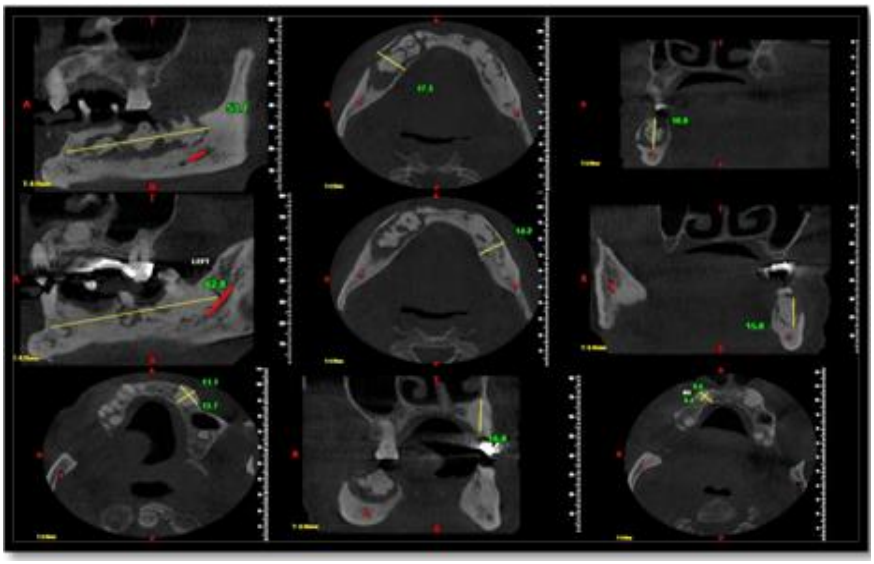

Figure 2: Cone-Beam Computed Tomography scan showing multifocal mixed radiolucent-radiopaque lesions

On both sides of the jaw, the lesion was approximately 52.8 mm x 18.6 mm x 17.3 mm in its greatest anteroposterior, supero-inferior, and medio-lateral dimensions. The internal structure was radiolucent and radiopaque, with well-defined sclerotic radiopacity within sclerotic radiolucent boundaries (Figure 3).

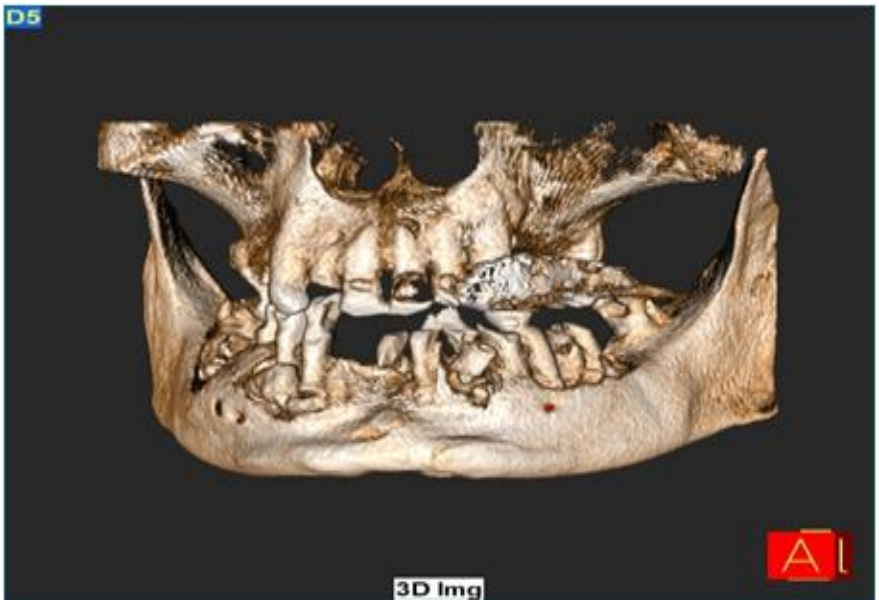

Figure 3: Three-dimensional reconstruction computed tomography image of the patient showing multifocal mixed radiolucent-radiopaque lesions

The borders were well defined, with noticeable bucco-lingual cortical expansion, thinning, and occasional effacement of cortices. The circular / oval radiopaque masses were continuous with the root apices in most locations. The basal maxilla had a well-defined radiopacity contiguous to the root apex of tooth 13 and 23, with entirely radiopaque internal structure and the opacity continuing till the apical alveolus. The greatest antero-posterior, mesio-lateral, and supero-inferior dimensions of the lesion are approximately 6.4 x 9.6 x 11.4 mm w.r.t. 13, and approx. 13.7 x 11.1 x 16.8 mm w.r.t. 23. Based on the radiographic findings, differential diagnosis of benign odontogenic tumor, fibroosseous lesion, and chronic osteomyelitis were considered. Patient was further advised for complete blood hemogram, HBA1C, serum calcium and serum alkaline phosphatase and the findings were in normal range with slightly elevated serum calcium and alkaline phosphate levels. Hence, on the basis of clinical, radiological and biochemical investigations, final diagnosis of FOCD was given.

Discussion

Classification of CODs has been a constant topic of debate since discovery of FCODs by Melrose et al. in 1976 who described these lesions as the entities involving multiple quadrants of jaw bones and they are thought to arise from constituents of periodontal ligament [3]. Plethora of terminologies have been suggested for the aforementioned group of lesions and few of them include sclerosing osteitis, gigantiform-cementoma, multiple cemento-ossifying fibromas, periapical cementoblastoma, and gigantiform-cementoma[1].

FCODs are situated in the periapical regions of teeth, and are mainly radiolucent (osteolytic phase), mixed (cementoblast phase), and finally turn radiopaque (osteogenic phase) with a thin radiolucent peripheral halo [4].

FCODs are characterised as non-expansile intraosseous masses of variable degree of lucency surrounding the root apices of vital teeth or edentulous areas in the posterior jaw [1]. Multifocal distribution and involvement of more than one quadrant of the jaws, typically in a bilateral symmetric fashion is pathognomonic for these lesions which was there in the present case.

Also, FCODs are not linked with any serum biochemical disturbances, skeletal disorders and systemic manifestations which can act as a guiding tool for distinguishing it from other similar entities [1]. Similarly, the present case revealed normal biochemical findings, hence confirming the diagnosis.

Diagnosis of FCODs based on demographic, clinical and imaging features are essential as the involved bone is more prone to infections due to invasive procedures such as biopsies. Henceforth, Cone Beam Computed Tomography (CBCT) scan can be a valuable tool for identifying such lesions, as it provides accurate assessment of the bone tissue. Differentiating FCOD from bone sclerosis with the help of aforementioned features are essential in cases of implant placement in such areas, as the presence of COD contraindicates this procedure [5]. On similar grounds, implant placement was avoided in the present case.

The alveolar bone of root apices of vital teeth manifests delayed healing because of the poor blood supply of the lesion, thereby increasing the occurrence of infection, ultimately leading to osteomyelitis of the jaws [6]. Hence, early diagnosis can play a preventive role in such scenarios.

Over the previous five years, a summary of

FCOD case reports has been presented. (Table 1)

Table 1: Literature Review

Author (year)	Demographic data	Clinical presentation	Radiographic features	Management
Cavalcante et al [7] (2016)	49 yr/F ;Right mandible	Facial asymmetry - scar and fistula and pus discharge. Intraoral examination - vestibular cortical bulging.	Sclerosis and osseous sequestrum	Complete bony fragment removal with reduction of mandible base
Daviet-Noual et al [8] (2017)	*Case 1*- 64yr/M African ;Left maxilla *Case 2*- 50yr/F African ;Left mandible *Case 3*- 72yr/F Cameroonian ;Left maxilla	*Case 1* - mobile and painful max. left first molar resulting from a periodontal abscess, *Case 2*- pain and tenderness on the lower left second molar, *Case 3*- multiple tooth pain, more severe in the upper left maxillary region.	*Case 1*- Multiple periapical radiolucencies, particularly mandibular body *Case 2*- mixed-density periapical lesion - round periapical radiopaque lesion (approx. 7mm in diameter) centred on the mesial root within a globally radiolucent periapical lesion encompassing both roots of the tooth. On panoramic radiography, similar lesions were seen across the jaw *Case 3*- several large radiolucent and radiopaque lesions in the periapical areas of all mandibular molars.	*Case 1*- No treatment required *Case 2*- Endodontic treatment *Case 3*- incisional biopsy of lesion confirming the diagnosis of FCOD.
Esfahanizadeh et al [9] (2018)	62-yr/ F Caucasian ;Mandible	Partial edentulous (bilateral posterior) mandibular	Radiolucent lesions in the periapical area of mandibular incisors. Two similar bilateral lesions in the mandibular molar area.	Insertion of two dental implants in the mandibular left edentulous area. Highly cautious surgery approach was taken as any surgical incision would have a devastating effect on FCOD lesions
Brooks et al [10] (2020)	66 yr/F ;Bilateral mandible	Bilateral mandibular florid cemento-osseous dysplasia, a compound odontoma next to the former site of the mandibular right canine (COD), Multiple hypercementosis sites, mild to severe hypercementosis periodontitis and apical periodontitis in the maxillary left first molar	Horizontally transmigrated mandibular right canine extending to apical area of the left first premolar along with bilateral tiny tonsilloliths and calcified stylohyoid ligaments	Surgical removal of the odontoma and canine teeth, followed by a biopsy to rule out cystic pathology.
Yakoob et al [2] (2020)	*Case 1*- 58yr/ F;Right maxilla *Case 2*- 18yr/ daughter ;mandible *Case 3*- 21yr/ grandson (familial) ;Anterior mandible	*Case 1*- facial asymmetry and a protrusive right maxilla for several years, *Case 2*- her mandible appeared expansive and protrusive. *Case 3*- expansive, protrusive mandible and associated malocclusion along with large intraoral swelling and ulcer	*Case 1*- extending irregular mixed radiolucent-radiopaque mass shifting the walls from the third molar to the midline. Multiple infections of the maxillary sinus, orbit, and nasal cavity *Case 2*- radiolucent-radiopaque mixture places all around the place Mandible and maxilla a number of teeth were affected, and an extra molar tooth was extracted the right maxillary third molar is coronal to this tooth. *Case 3*- well-demarcated, cortication loss on the superior aspect. mixed diffuse radiolucent-radiopaque throngs in each of the four quadrants with a large number of impacted teeth	*Case 1*- The necrotic bone in the right maxilla was surgically debrided and sent for histopathologic examination, which revealed a calcified mass with globules of cementum-like material, acute osteomyelitis bone, and actinomyces bacterial colonies *Case 2*- the patient was referred to the orthodontic department for malocclusion treatment *Case 3*-The diagnosis of ossifying fibromatoid lesion" occurring in a background of familial florid COD was established by incisional biopsy, and the patient was thereafter lost to follow-up.
Panta et al [11] (2021)	*Case 1*- 30yr/F; Mandible *Case 2*- 50yr/F;Left mandible	*Case 1*- a number of - permanent teeth that have been completely destroyed *Case 2*-nothing relevant	*Case 1*- Multiple radiopacities in the mandible's right premolar-molar area and weakly calcified radiopacities on the left side, as well as a strange trabecular pattern. *Case 2*- #35, #36 & #37 and #45, #46 (mesial, and distal root tip) of the mandible, well-defined radiopaque lesions with a radiolucent halo.	*Case 1*- Extraction was advised *Case 2*- Endodontic treatment with follow up
Thakur et al [12] (2021)	40yr/ F Left mandible	Nothing abnormal	44, 45, 47, and 35 have a multifocal radiopaque mass in the periapical region.	Biopsy confirmed diagnosis of FOD

It has been highly recommended that the large FCOD lesions require complete surgical removal, since recontouring can lead to significant regrowth. However, the requirement of surgical intervention for improved function should be evaluated for each case, and should be assessed for any risk of introducing infection in the lesional a vascular bone [2].

Conclusion

Since the diagnosis of FCOD is mainly based on the clinical and radiological features; biopsy must be avoided because of the fact that inoculation with oral microbes may precipitate chronic infection in such hypovascular lesions.

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