Case Report

Florid cemento-osseous dysplasia in a young Indian female: A rare case report with review of literature

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Abstract

Florid cemento-osseous dysplasia (FCOD) is a reactive bone dysplasia that involves tooth-bearing areas of the jaws. It most commonly affects females and is seen less frequently in Caucasian and Asian groups. The age group may vary from 19 to 76 years and typically presents in the fourth and fifth decades. FCOD associated with impacted teeth and bony expansion is very rare and only a few familial cases have been reported in literature. Till date, <2% cases in Indians have been reported in literature. Here, we report a case of nonfamilial FCOD in a 27–year-old young Indian female with cortical plate expansion, one over-retained and one impacted teeth.

Keywords: Cemento-osseous dysplasia, florid cemento-osseous dysplasia, impacted teeth, over-retained teeth

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INTRODUCTION

Florid cemento-osseous dysplasia (FCOD) is a benign, multifocal, fibro-osseous lesion of the jawbones consisting of cellular fibrous connective tissue along with bone- and cementum-like tissues.[1] FCOD was previously known by various names such as gigantiform cementoma, multiple cemento-ossifying fibroma, sclerosing osteitis, multiple enostosis and sclerotic cemental masses of the jaws.[2] The term FCOD was first described in 1976 by Sentürk *et al.* to describe a condition of exuberant multiquadrant masses of cementum and/or bone in both the jaws, and in some cases, simple bone cavity-like lesions in the affected quadrant.[3,4] It is usually asymptomatic and often discovered as an incidental radiographic finding; however, it may cause facial deformity, resulting from the displacement of cortical plates. In some cases, dull pain or pus discharge may occur usually associated with exposure of sclerotic calcified masses in the oral cavity.[4] In this case report, we present a case of nonfamilial FCOD in a 27-year-old young Indian female with cortical plates expansion, one over-retained tooth and impacted tooth.

CASE REPORT

A 27-year-old female patient has been reported to the department of oral medicine and radiology with the chief complaint of swelling in the lower right front teeth region of the jaw for 2 years. The patient's history

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revealed that initially the swelling was smaller in size and has increased gradually to the present size. The swelling was not associated with pain or any other symptom. The patient's past dental history revealed the extraction of left maxillary posterior tooth 8 months back due to caries and the healing was uneventful. On extraoral examination, single localized swelling with diffuse margins of size approximately 2 cm × 2 cm was present on lower one-third of the right side of the face below the corner of the mouth with normal-appearing overlying skin [Figure 1a and b]. On palpation, the swelling was afebrile, nontender and firm to hard in consistency. On intraoral examination, single diffuse swelling of size approximately 1 cm × 2 cm was present on the lower right buccal vestibule in relation with 43 and 44 regions, obliterating the buccal vestibule [Figure 2]. The swelling was nontender and hard in consistency. Right permanent maxillary first molar was missing, left mandibular canine and second deciduous molar were over retained and left mandibular permanent canine and second premolar were clinically missing. On the basis of history and clinical examination, a provisional diagnosis of central ossifying fibroma was given. Orthopantomograph (OPG) was advised which revealed multiple bilateral radiopaque sclerotic lesions surrounded by peripheral radiolucent rim apical to majority of the mandibular teeth, with no evidence of root resorption and impacted left mandibular permanent canine and second premolar [Figure 3]. Computed tomography (CT) scan revealed the presence of an expansile lytic lesion in the right side body of the mandible measuring 3.5 cm × 2.9 cm with hyperdense matrix and intact cortex and a small lesion on the left side body of the mandible with a connection in between [Figure 4a and b]. The biochemical investigations were advised which showed increased alkaline phosphatase levels. Thus, a working diagnosis of FCOD was given. Incisional biopsy was taken from the lower right buccal vestibule region and was sent for histopathological examination. The histopathologic section showed numerous woven bony trabeculae in a fibrocellular stroma with numerous ossicles and few cementicle-like areas. Stromal cells were spindle-shaped and arranged in haphazard manner. Osteocytes and rimming osteoblasts were evident [Figure 5]. The histopathological picture was suggestive of cemento-osseous dysplasia (COD). Thus, the final diagnosis of FCOD was confirmed. The treatment done was facial recontouring under local

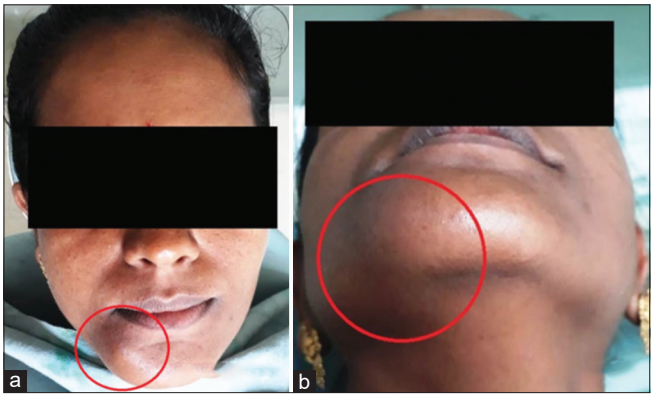

Figure 1: (a-b) Extraoral picture of the patient with evident swelling in the right side parasymphysis region

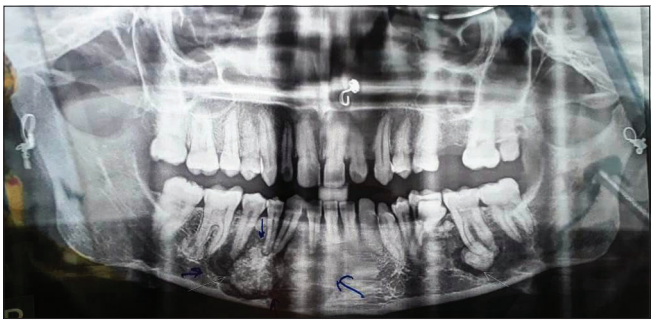

Figure 3: Orthopantomograph of the patient showing multiple bilateral radiopaque sclerotic lesions surrounded by peripheral radiolucent rim apical to majority of the mandibular teeth

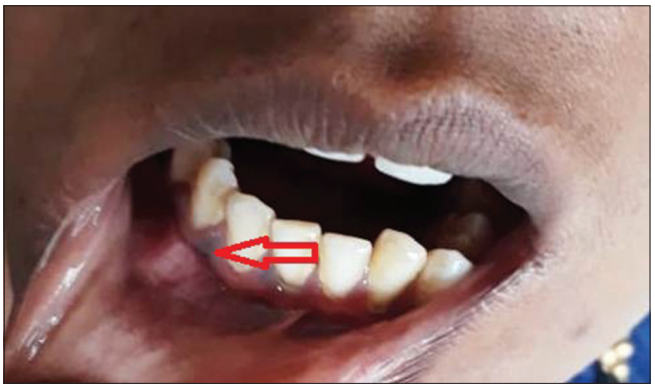

Figure 2: Intraoral picture of the patient showing vestibular swelling in 43 and 44 regions

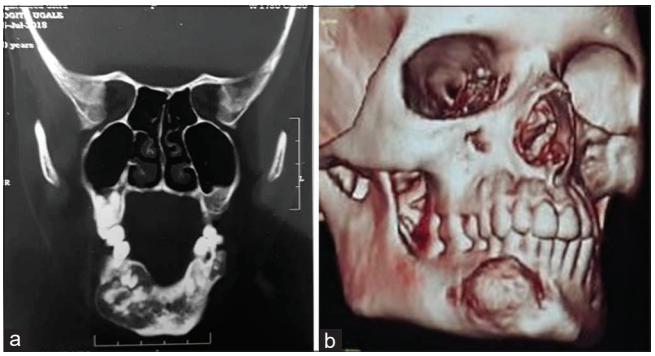

Figure 4: (a) Computed tomography scan image showing expansile lytic lesion with hyperdense matrix and intact cortex involving almost whole of the mandible. (b) Three-dimensional reconstruction computed tomography image of the patient showing bony overgrowth in the right mandibular parasymphysis region

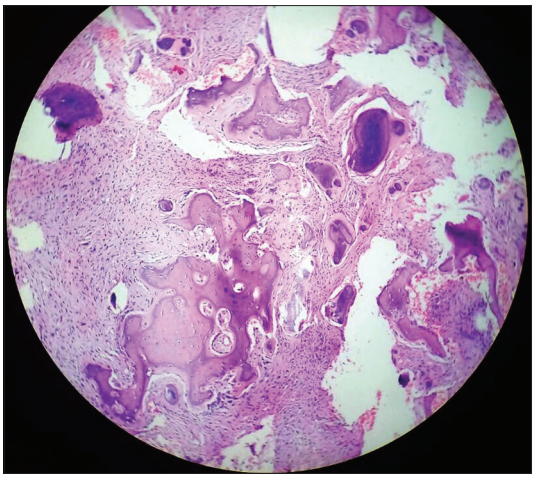

Figure 5: Histopathological picture showing numerous woven bony trabeculae in a fibrocellular stroma with numerous ossicles and few cementicle-like areas

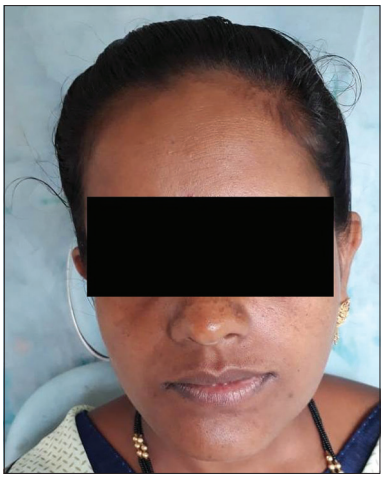

Figure 6: Postoperative follow-up picture of the patient after 1 month

anesthesia, and the patient is on regular follow-up with no signs of recurrence [Figure 6].

DISCUSSION

The current classification of cementomatous lesions by the World Health Organization (WHO) published in 2005 is based on age; sex; histopathological, radiographical and clinical characteristics and location of the lesion. This classification includes the following three groups: cemento-ossifying fibroma, benign cementoblastoma and COD.[5] COD is a group of disorders known to originate due to reactive or dysplastic changes in the periodontal ligament tissues (Waldron *et al*.).[6,7] They are usually classified, depending on their extent and radiographic appearances, into three main types: periapical CODs (surrounding the periapical region of teeth bilaterally), FCODs (sclerotic symmetrical masses involving more than one quadrant) and FCODs (single lesion).[4,7] The term FCOD has been proposed in the second edition of the WHO's "International histological classification of odontogenic tumours" to replace the first edition's "gigantiform cementoma."[6] The word "florid" refers to the widespread and extensive manifestations of the disease in the jaws.[7,8]

FCOD is a reactive process with the involvement of tooth-bearing areas of the jaws.[7] Clinically, FCOD is asymptomatic, but sometimes, there may be localized expansion of the cortical plates, as was seen in the present case or symptoms of dull aching pain or drainage may be present. FCOD typically occurs in middle-aged black women.[7] Melrose *et al*. reported 34 cases of similar lesions, of which 32 were black women (in predominantly Caucasian population) with a mean age of 42 years.[3] However, in the present case, the patient was 27 years female.

MacDonald-Jankowski published a systematic review on FCOD in 2003, regarding the comparison of occurrence of FCOD in oriental and nonoriental population, in which he reviewed a series of 156 females who were observed in 17 series of patients. Among them, 59.6% (93 patients) were dark- skinned, 37.2% (58 patients) were oriental and 3.2% (5 patients) were Caucasians and Indians. Only three Indian patients were identified from the whole series that combined most of the cases reported in literature around the world which makes the occurrence of FCOD to be <2% in the Indian population. The same systematic review study found that the mean age for those patients was 47–49 years.[6] However, we reported a rare case of FCOD in a 27-year-old young Indian female. Bagheri *et al*.[7] in 2014 reported two cases of FCOD in 34-year- and 36-year-old females and one case in 61-year-old female. Sentürk *et al*. in 2013 reported the first case of FCOD in a 35-year-old Turkish male.[3]

Familial FCOD is quite uncommon, and there are only a few cases reported in literature where the hereditary nature of the lesion could be demonstrated. The familial form tends to occur in younger individuals and is characterized by more expansile lesions that may recur after surgery. In all of the familial cases reported in literature, FCOD appears to be inherited as an autosomal dominant trait with variable phenotypic expression. Toffanin *et al*. reported a case of familial FCOD with multiple impacted teeth and marked expansion of the symphyseal region. Multiple impacted teeth are a rare phenomenon in cases diagnosed as FCOD. Srivastava *et al*. also reported a case of familial FCOD associated with multiple impacted teeth.[3] However, in the present case, we have reported one impacted tooth and one over-retained tooth in association with nonfamilial FCOD, which is a rare finding.

The radiographic appearance of FCOD depends on its degree of maturation. The lesion may appear as radiolucent, mixed or lobulated dense radiopaque masses with radiolucent rim, usually located in tooth-bearing areas.[2] Most cases occur bilaterally in the mandible and usually with no cortical expansion or pain.[7] However, in the present case, there was cortical expansion on the right side of the body of the mandible. OPG in the present case also showed multiple bilateral radiopaque sclerotic lesions surrounded by peripheral radiolucent rim apical to majority of the mandibular teeth. Most of the times, these lesions are diagnosed incidentally on routine radiographic examination. However, in the present case, it was not an incidental finding. Other than routine radiographs, axial and cross-sectional CT and cone-beam computed tomography images clearly show the location and extent of the lesion, expansion of the cortical bones which helps to differentiate FCOD from lesions that exhibit a similar sclerotic appearance on conventional radiographs.[2] In the present case also, CT scan was advised which showed an expansile lytic lesion with hyperdense matrix and intact cortex.

The WHO report of 1992 describes CODs as a variety of jaw lesions that are characterized histologically by the presence of cementum-like tissue and which appear to be dysplasias rather than neoplasms.[8] Similarly, in the present case, the histopathologic section showed numerous woven bony trabeculae in a fibrocellular stroma with numerous ossicles and few cementicle-like areas.

Often, differential diagnosis plays a crucial role, especially when there is coincidental finding such as odontogenic infection or neoplasia and bone dysplasia that can cause changes in the mandible with similar radiographic characteristics. Hence, all the available diagnostic information should be considered which will lead to an adequate diagnosis.[4] Some other lesions with similar features such as FCOD are considered in the differential diagnosis including Gardner's syndrome, Paget's disease, chronic diffuse sclerosing osteomyelitis, cemento-ossifying fibroma, osteoblastoma, odontoma, ameloblastic fibro-odontoma, osteoid osteoma, calcifying epithelial odontogenic tumor (CEOT), ossifying fibroma and fibrous dysplasia. Unlike Gardener's syndrome, FCOD does not present other skeletal changes, no skin tumors or dental anomalies and thus can be easily differentiated. Paget's disease affects most commonly white males and is polyostotic with pathognomonic increase in serum alkaline phosphatase level which is not a consistent feature of FCOD. However, in the present case, increased alkaline phosphatase levels were found. Chronic diffuse sclerosing osteomyelitis is generally unilateral with soft-tissue swelling, fever and lymphadenopathy affecting primarily the mandible with cyclic episodes of pain and is not always limited to the tooth-bearing areas. The affected region of the mandible exhibits a diffuse opacity with poorly defined borders. Sometimes, it may be the complication of FCOD. Cemento-ossifying fibroma exhibits more buccolingual expansion than FCOD, whereas osteoblastoma causes more expansion of bony plates and has radiolucent rim.[6] Odontoma and ameloblastic fibro-odontoma generally occur in children and are in association with the impacted teeth. Osteoid osteoma and osteoblastoma usually occur during the second decade of life, and the major symptom associated is the dull nocturnal pain that FCOD lacks. CEOT, ossifying fibroma, and fibrous dysplasia are benign, slow-growing expansile lesions, while FCOD rarely shows bone expansion.[1,2,4-6]

The diagnosis of FCOD is principally based on clinical findings, localization of the lesion, patient's age, gender and ethnicity as well as radiological features.[9] As such, biopsy is not required in these cases since the lesion can be diagnosed radiographically. Instead, follow-up and recontouring can be recommended when cortical expansion occurs, as was done in the present case. Since surgery may lead to the lack of vascularity of the lesion and increased the risk of osteomyelitis of the bone, it is not normally justified to surgically remove these lesions. The affected area undergoes changes from normal vascular bone into avascular cementum-like masses. Furthermore, complete removal of necrotic tissue may result in large discontinuity defect which ultimately results in patient morbidity. In asymptomatic patients, recommended treatment is observation and regular radiological follow-up. However, in those lesions which cause pain and disturbance, surgery as well as the associated risks should be considered for adequate treatment. Surgical recontouring should be the treatment of choice in cases where there is only cortical expansion with or without mucosal perforation due to the cemento-osseous lesions, as was done in the present case.[1,2]

CONCLUSION

Diagnosis of pathologies in the jaw is based on clinical findings, radiographic features and histological picture. However, FCOD can be diagnosed based on its clinical and radiographical features. Although not mandatory, biopsy may be done to confirm the diagnosis. Bone expansion and impacted teeth are very rarely seen in nonfamilial forms of FCOD, which makes this case rare. If the lesions

and impacted teeth are asymptomatic, it is better to avoid surgical intervention. However, in the present case, due to esthetic reasons, facial recontouring was done.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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