Case Report

Florid cemento-osseous dysplasia: A rare case report evaluated with cone-beam computed tomography

Eren Yildirim, Serdar Bağlar[1], Mehmet Ertugrul Ciftci[2], Erdal Ozcan[3]

Department of Maxillofacial Radiology and [1]Department of Restorative Dentistry, Kirikkale University, Kirikkale, Faculty of Dentistry, [2]Department of Maxillofacial Radiology and [3]Department of Endodontics, Gaziantep University, Gaziantep, Turkey

Abstract

A 29-year-old systemically healthy female patient presented to our department. Cone-beam computed tomographic images showed multiple well-defined sclerotic masses with radiolucent border in both right and left molar regions of the mandible. These sclerotic masses were surrounded by a thin radiolucent border. We diagnosed the present pathology as florid cemento-osseous dysplasia and decided to follow the patient without taking biopsy. For the patient, who did not have any clinical complaints, radiographic followupis recommended twice a year. The responsibility of the dentist is to ensure the follow-up of the diagnosed patients and take necessary measures for preventing the infections.

Key Words: Bone disease, cone-beam computed tomography, florid cemento-osseous dysplasia.

Address for correspondence:
Dr. Eren Yildirim, Department of Oral and Maxillofacial Radiology, Faculty of Dentistry, Kirikkale University, Kirikkale, Turkey.
E-mail: dt.eren@hotmail.com
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INTRODUCTION

Current classification of cementomatous lesions published in 2005 by the World Health Organization (WHO) is based on age, sex and histopathologic, radiographic and clinical characteristics, as well as the location of the lesion. This classification includes cemento-ossifying fibroma, benign cementoblastoma and cemento-osseous dysplasia (COD) groups.[1] Florid COD (FCOD) is one of the subgroups of COD. It was previously known as gigantiform cementoma, multiple cemento-ossifying fibroma, sclerosing osteitis, multiple enostosis and sclerotic cemental masses of the jaws. FCOD was first described by Melrose *et al.* in 1976.[2]

This lesion is most commonly found in middle-aged dark-skinned women although it may also occur in Caucasians and Asians. Since FCOD lesions are seen close to periodontal ligament and have similar histopathology, it has been suggested that origin and pathogenesis of these lesions is from periodontal ligament; therefore, few authors have reported that the remains of cementum in bone after extraction might be a reason for FCOD. However, the exact etiology of FCOD is still unknown.[3-5]

Clinically, FCOD may be asymptomatic, and in such cases, the lesion is incidentally detected during routine radiographic examination. In severe cases, where infection occurs, dull pain, drainage, exposure of the lesion in oral cavity, focal expansion and facial deformities are present.[6,7]

Histopathologically, FCOD shows irregular shaped, dense, cell-free cemental masses and nonlamellar bone masses in fibroblastic connective tissue.[3-7]

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Radiographically, FCOD is characterized by multiple masses of mixed radiopaque strucutres. Often with a circumferential radiolucency, primarily surrounding the root apices of vital teeth, and over time with the maturation of the lesions, radiographic images can become increasingly radiopaque. These lesions are most commonly seen symmetrically in mandibular premolar-molar regions and are often confined within the alveolar bone, and can be seen associated with maxillary regions.[1,2,6,7]

In asymptomatic cases, diagnosis can be made based on these radiographic presentations; however, differential diagnoses is important to eliminate the other lesions such as Paget's disease, gigantiform cementoma, chronic diffuse sclerosing osteomyelitis, fibrous dysplasia, periapical cemental dysplasia and osteosarcoma that may have similar radiographic appearances with FCOD.[1-3,6-9] In these aspects, computed tomography (CT), because of its ability to give axial, sagittal and frontal views, is useful in the evaluation of these lesions.[7,10]

In the management of these conditions clinical and radiographic follow-up must be the first step. Treatment is required when infection, pain or drainage of the lesion occurs.[8]

In this paper, a case of a patient was presented who was diagnosed with FCOD on the basis of clinical, radiographic and biochemical findings.

CASE REPORT

A 29-year-old light-skinned female patient was referred to Faculty of Dentistry, for the treatment of her cracked and missing teeth. She had no medical problem and also no unusual situation could be determined during clinical extra- and intra-oral examination. Panoramic radiography was performed to evaluate the dental arches and ovoid radiopaque masses in radiolucent spaces were detected in relation to both mandibular left and right molar regions under fixed partial dentures [Figure 1]. Lesions were symmetrical. For a more detailed assessment, cone-beam CT (CBCT) was taken, and multiple well-defined sclerotic masses surrounded by a thin radiolucent border were seen more clearly in the cross-sectional images [Figures 2-5]. Axial sections of 1 mm thickness were obtained. The CBCT scan showed bone lesions enlarged in the alveolar bone, but no cortical plate expansion was observed. Superior border of mandibular canal was intact. Slight thinning of the cortical plate in the regions was noticed (especially at the left mandibular buccal side). Based on all these features, a final diagnosis of FCOD was made, and the differential diagnosis of chronic sclerosing osteomyelitis and Paget's disease was considered. Biochemical analysis of serum alkaline phosphatase, calcium and phosphorus was carried out to differentiate from Paget's disease and was found within normal limits.

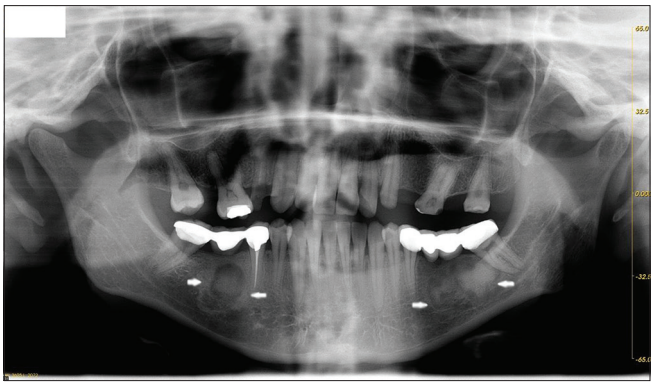

Figure 1: Orthopantomography showing multiple ovoid radiopaque masses in the mandibular molar area bilaterally

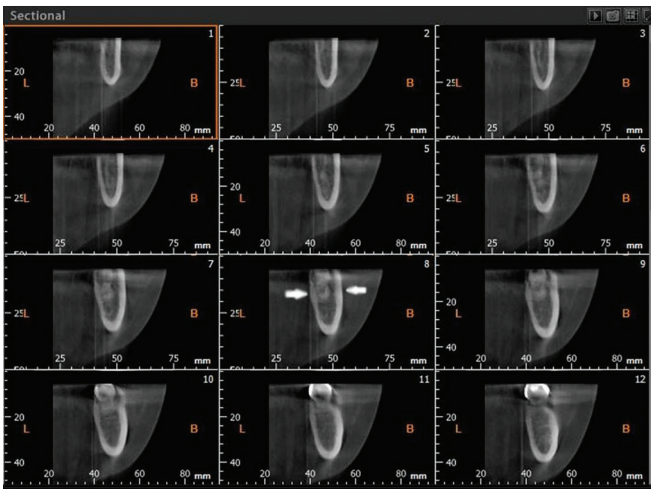

Figure 2: Cross-sectional CBCT images of right mandibular molar region showing slight thinning of the cortical plate but no expansion in the buccal or lingual cortical bone

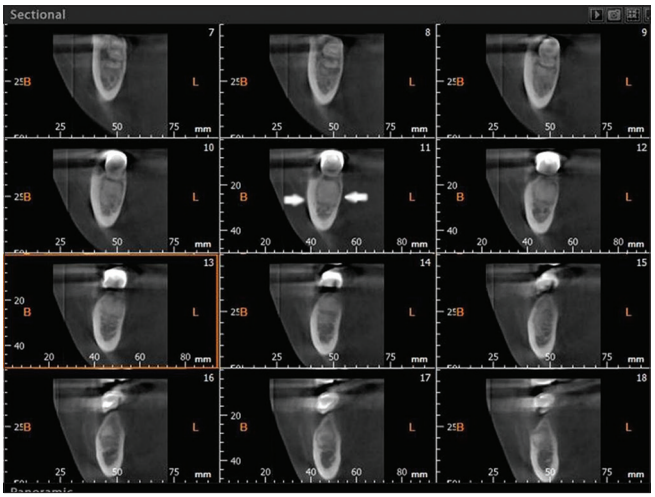

Figure 3: Cross sectional CBCT images of left mandibular molar area revealed slight thinning of the cortical plate but no expansion in the buccal or lingual cortical bone

Biopsy was not done due to the avascular nature of the lesion which contributes to susceptibility of the lesion to severe infection,

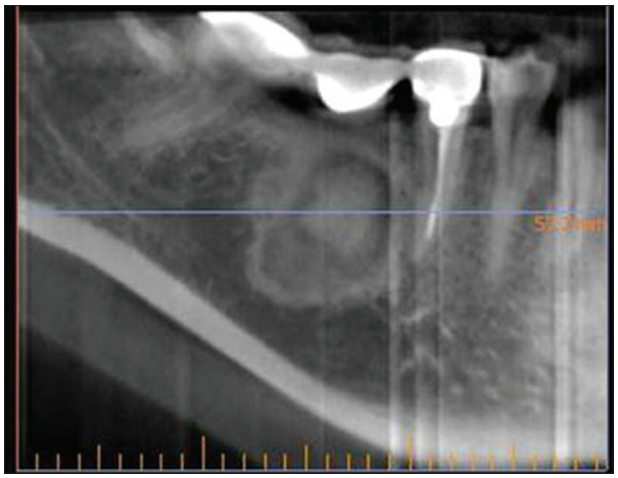

Figure 4: Closer view of the right mandibular molar region on sagittal cone-beam computed tomography image revealed radiopaque lesion covered with radiolucent band

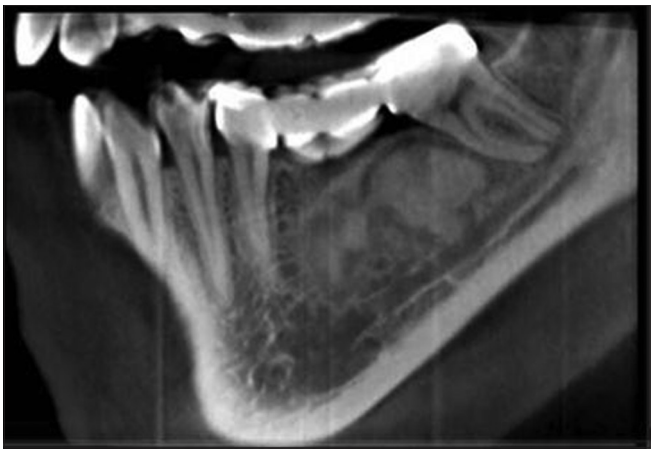

Figure 5: Closer view of left mandibular molar region on sagittal cone-beam computed tomography image revealed radiopaque lesion covered with radiolucent band

bone sequestration and osteomyelitis when surgery is performed. The treatment plan included good oral hygiene maintenance and periodic follow-up. Surgical intervention was contraindicated for this case because of the asymptomatic presentation of the lesion.

DISCUSSION

In 2005, the WHO classified the bone-related lesions, "COD" is one of the groups of this classification, and FCOD is one of the subgroups of COD which is a non-neoplastic fibro-osseous lesion.[1,3] Focal COD and periapical COD are the other COD lesions. The differentiation of these lesions is based on clinical characteristics, location and radiographic features such as localization and diffuseness.[1,3,4]

The etiopathogenesis of FCOD is not clear but reactive or dysplastic changes in the periodontal ligament might be a cause for the disease. It is important to consider the clinical, radiographical and histopathological differential diagnosis of FCOD and to differentiate from other lesions such as periapical cemental dysplasia, Paget's disease, Gardner's syndrome, chronic diffuse osteomyelitis, cemento-ossifying fibroma and fibrous dysplasia. Periapical cemental dysplasia is often seen at apices of anterior teeth and usually does not exceed a limit of 1 cm. Focal cemento-osseous dysplasia is often seen at two or more mandibular anterior teeth or at the apices of molar region, and does not grow more than 2 cm, FCOD appears bilaterally, and mostly in the mandibula and often presents symmetrically. Radiolucent band surrounding the radiopaque lesion can be seen or the lesion can be completely radiopaque.[11,12] FCOD is a nonneoplastic, reactive process which is most frequently reported in middle-aged women of African descent, but the definite female gender predilection cannot be explained. Paget's disease of bone may mimic FCOD on radiological evaluations; the difference is that FCOD is seen above the inferior alveolar canal, whereas Paget's involves the entire mandible and exhibits loss of lamina dura. In this aspect, CBCT can be used in differentiation of FCOD from other lesions which have similar sclerotic appearance on conventional radiographs because it gives the clear view of axial aspect of the lesion. In addition, biochemical serum changes such as elevated alkaline phosphatase levels occur in Paget's. In our case, CBCT scan evaluations showed that the inferior alveolar canal was not affected, and there was no serum alkaline phosphatase level change. Gardner's syndrome also presents with other skeletal changes, skin tumors and dental anomalies, but FCOD is only seen at tooth-bearing areas. Differentiation from chronic diffuse osteomyelitis is also similar because chronic diffuse sclerosing osteomyelitis involves the body of the mandible from the alveolus to the inferior border and may extend into the ramus, and it appears as single, poorly delineated opaque segment of the mandible, whereas FCOD is seen as multiple round or lobulated opaque masses only at tooth-bearing areas. Cemento-ossifying fibroma which is a neoplastic lesion, displays more severe buccolingual expansion than does FCOD since it is not a neoplasia. Fibrous dysplasia has pathognomonic ground glass appearance in radiologic images; additionally, FCOD is not a developmental lesion unlike fibrous dysplasia.

A 29-year-old female patient who was diagnosed on routine radiographic examinations is presented in this report.

Three radiographic images of developmental stages of FCOD are:

- Osteolytic stage is the initial stage of the lesion, shows loss of lamina dura and periodontal ligament, and is seen as well-defined radiolucent areas
- In cementoblastic stage, radiopaque zones appear in radiolucent area. These areas are composed of cementum-like calcified structure in fibrous tissue

- Last stage is the mature stage seen as definite radiopaque masses in majority of the lesion.[12,13]

In the present case, all types of the radiographic views could be detected. The left lesional area showed cementoblastic and mature radiographic images, the right lesional area showed cementoblastic image in relation to the bigger lesion and under this lesion, osteolytic appearance was present.

Usually, clinical and radiographic examinations and findings would be sufficient to diagnose FCOD. Furthermore, CBCT imaging should be considered if new symptoms or signs develop.[10,13] To the best of our knowledge, a few FCOD cases diagnosed with CBCT have been reported in the literature; furthermore, CBCT images are useful diagnostic tools for identifying the location and extent of the lesion.[14]

In such asymptomatic cases, biopsy for histological examination is not recommended because of risks of infection, sequestrum formation and osteomyelitis.[3,5,10-13] In this kind of asymptomatic situations, the best choice is routine radiological and clinical follow-ups and maintenance of the oral hygiene status to prevent possible infections. If the case becomes symptomatic, antibiotic therapy and sequestrectomy should be done

In this recent asymptomatic case, we have diagnosed the lesions as FCOD based on the clinical and characteristic radiographic features. Furthermore, CBCT image analysis were made in addition to panoramic radiographs, to support the diagnosis. The expansion of the cortical bones was clearly evaluated on CBCT, even if it was slight. Besides, we prefer not to do histological examination because of above-mentioned risks.

It is of major importance that dentists should be very careful in such cases because this kind of radiolucencies may be misdiagnosed or overlooked and this may cause unnecessary endodontic or surgical treatments that may lead to complications.

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Conflicts of interest
There are no conflicts of interest.

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