

Original Article

Florid osseous dysplasia coexisting with focal osseous dysplasia in the jaw: a case report and literature review

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Abstract: Cemento-osseous dysplasia (COD) is a disease characterized by the replacement of normal bone with fibrous tissue and metaplastic bone, and occurs in the tooth-bearing areas of the jaws. It is usually asymptomatic and often found incidentally on radiographs. Herein, we report a rare case in which florid COD (FLCOD) and focal COD (FOCOD), two different forms of COD, coexisted in the maxilla of a Chinese female patient. FOCOD was found in the left maxilla, and FLCOD was located in the three other quadrants of the jaws. The lesions were discovered when the patient underwent treatment for a secondary infection. The findings from this study may aid in the recognition, diagnosis, and treatment of COD.

Keywords: Cemento-osseous dysplasia, florid osseous dysplasia, focal osseous dysplasia

Introduction

Florid cemento-osseous dysplasia (FLCOD) is a benign lesion affecting the jaws and was first reported by Melrose et al. in 1976 [1]. The etiology of this condition is unclear; however, it is generally considered to originate from the periodontal ligament [2-4]. FLCOD occurs most often in middle-aged black women, although it may also occur in Asians and Caucasians [5, 6]. Patients with FLCOD generally present with no obvious clinical symptoms as the related teeth are often vital [7-9]; therefore, the condition is often discovered during routine X-ray examinations. Sometimes patients present with local swelling, pus, and pain in the affected area following a secondary infection [5, 10]. Among the current diagnostic methods used, histopathology and radiography have proved to be useful for the diagnosis of FLCOD [6, 11]. The lesions are usually benign and require no treatment; however, anti-inflammatory drugs and surgery may be required in cases of secondary infection [2, 11, 12].

Focal cemento-osseous dysplasia (FOCOD) mainly occurs in females and always presents as a solitary lesion with single site involvement.

Although it may occur anywhere in the jaws, the posterior part of the mandible is the site most commonly affected. The disease is typically asymptomatic and detected only on radiographic examination as an irregular, mixed radiolucent-opaque lesion, surrounded by well-defined borders [13]. Herein, we present a rare case of cemento-osseous dysplasia (COD) involving all four quadrants of the jaw bone. Both FLCOD and FOCOD were found to coexist in different areas within the jaws of a single patient.

Case report

In January 2015, a 68-year-old Chinese female patient was referred to our hospital, with chief complaints of swelling, pus discharge, and pain in the right maxilla for the previous 1 month. She had been diagnosed with osteomyelitis and had undergone surgery involving curettage for a total of four times during the past 12 years. Examination of the panoramic radiograph and cone beam computed tomography (CBCT) scan showed the presence of several radiolucencies and radio-opacities in every quadrant of the jaws, except for the upper left quadrant where the alveolar bone was found to be absorbed subsequent to tooth extraction.



Figure 1. Panoramic radiograph of the patient showing multiple sclerotic masses with radiolucent areas distributed symmetrically in the maxilla and mandible.

The irregularly shaped, extensive lesions were mainly located at the apical areas of teeth 16, 17, and 37-47 and appeared to blend with adjacent normal bone. Intact periodontal ligament spaces were observed between the lesions and the corresponding teeth. In addition, a well-defined radio-opacity with a thin, slightly irregular, peripheral radiolucent rim was observed around the apical tip of tooth 28 (**Figures 1, 2**).

The patient did not suffer from any other systematic disease that may have had a bearing upon the condition. Intraoral examination revealed that part of the left maxilla was missing as a result of a previously performed surgery. Gingival redness and swelling was observed extending from the distal part of the left maxillary tuberosity to the midline, along with exposure of the necrotic, yellowish-white bone in the right maxilla (**Figure 3**). The inflamed tissues and bone were exposed under general anesthesia. Fragile, yellow, irregular bone pieces with small holes (honeycomb-like appearance) were seen. The boundaries between pathological and normal bone were unclear. Curettage was performed in the area involving teeth 11-17. Furthermore, with the patient's consent, a specimen of the bone located on the distal part of tooth 36 was obtained for further histological examination.

Specimens from both the right maxilla and left mandible showed similar histopathological changes. Pale yellow, irregularly shaped lesions were observed. Decalcified sections of the tissues were prepared, and woven as well as lamellar bone along with sclerotic masses of cementum-like material (SMCM) containing

less fibrous connective tissue were visible under microscope. The SMCM were disorganized and irregularly shaped like ginger roots. Parts of the SMCM were fused together with a few cells within; the borders of the SMCM were relatively regular with no obvious osteoblastic and osteogenic reaction. Small stove-shaped blood-sinus like structures could be seen among the local sclerotic masses (**Figure 4**).

Based on the clinical, histological, and radiographic findings, a final diagnosis of COD with secondary infection was reached.

Discussion

According to the current classification of odontogenic tumors released in 2005 by the World Health Organization, COD is an idiopathic condition localized in the periapical regions within the tooth-bearing areas of the jaws and are characterized by the replacement of normal bone by fibrous tissue and metaplastic bone. There are four types of COD based on the region of the lesion: periapical COD (PCOD) occurs in the anterior mandible and involves only a few adjacent teeth, whereas FOCOD occurs in the posterior quadrants of jaws. The two more extensive types of COD occur either bilaterally or in all four quadrants of the jaws [14]. One of them is FLCOD, which affects mainly middle-aged black women, whereas the other type, named familial gigantiform cementoma, is seen mostly in young people. Familial gigantiform cementoma causes obvious bone expansion and is an autosomal dominant hereditary disease. Both FLCOD and familial gigantiform cementoma are very rare forms of COD [10]. In the present study, FLCOD was found to involve the jaw bones in an asymmetric manner. No FLCOD was detected in the left maxilla where a major part of the quadrant was edentulous. However, FOCOD was located at the apical end of tooth 28. To the best of our knowledge, this is the first report of the occurrence of FLCOD and FOCOD in the maxilla of the same patient.

Radiography is the main method used to diagnose COD. Limited forms of COD (PCOD and

A rare case of COD

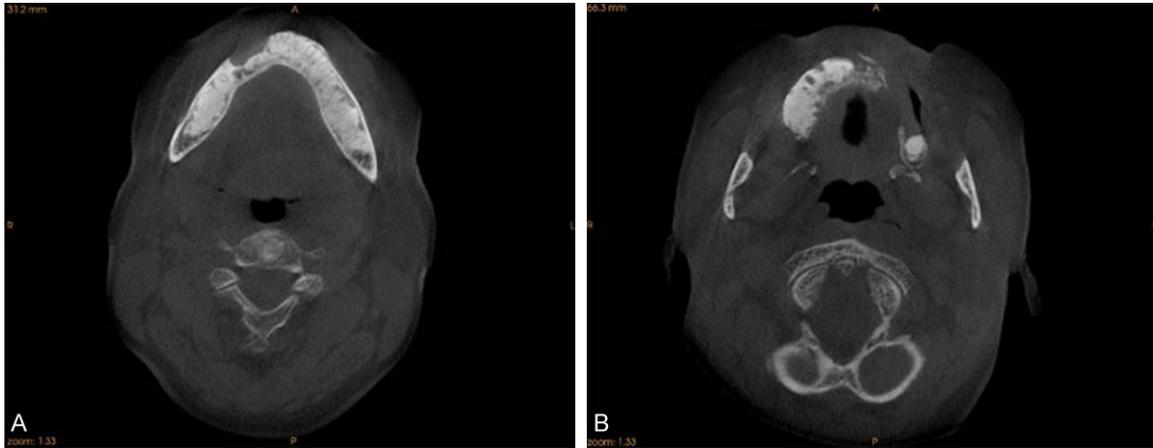


Figure 2. CBCT scan showing extensive radiopaque areas in the periapical regions on the right maxilla and both sides of the mandible. A focal lesion is seen around the tip of tooth 28.



Figure 3. Secondary chronic osteomyelitis of the right maxilla.

FOCOD) present as round or rounded lesions with clear boundaries (radiolucent thin rim around the lesion). The extensive COD types are mostly irregular in shape with undefined edges. The lesions typically demonstrate identical patterns of maturation during different stages. Initially, they present as predominantly radiolucent entities, with some areas of FLCOD showing low CT density and demonstrating the ability to develop into simple bone cysts [15]. Gradually, the lesions have a mixed followed by predominantly radiopaque appearance surrounded by a thin peripheral radiolucent rim. In the present case, the lesions were fused with each other and spread across the mandible. At the site of tooth 37, the lesion was found to extend beyond the mandible above the level of the alveolar ridge. Thus, there appears to be no relationship between the existence of a tooth and the progress of the lesion.

Histopathologically, COD lesions are composed of fibrous tissue and various types of cells. Most of them comprise woven as well as lamellar bone and SMCM. In most cases, the lesions do not fuse with the corresponding tooth roots but can blend with the adjacent normal-appearing bone, making it appear as if the lesion has no clear boundary. The amount of fibrous tissue decreases with time, whereas mineralization increases followed by the curving of the trabecular bone structure as the lesions progress, resulting in increased radio-opacity on X-ray [16].

As is often the case, COD lesions are usually asymptomatic and are found incidentally during routine radiographic check-up. Therefore, unnecessary dental pulp treatment can be avoided with the accurate diagnosis of these lesions. Most patients present at the hospital with swelling and pain caused by an infection secondary to the lesions. FLCOD cases are prone to infection due to denture pressure or a poorly healed extraction socket, leading to progressive atrophy of the alveolar bone and a lack of local blood supply. Most COD cases involve benign diseases with no serious outcomes. In general, unless the sclerotic bone in the lesions is infected (which is always in the case for FLCOD) or the lesion causes a facial deformity (as in the case of familial gigantiform cementoma), it does not require any treatment.

It is important to differentiate COD from other diseases, such as ossifying fibroma (OF), fibrous dysplasia (FD), chronic osseous sclerosis-

A rare case of COD

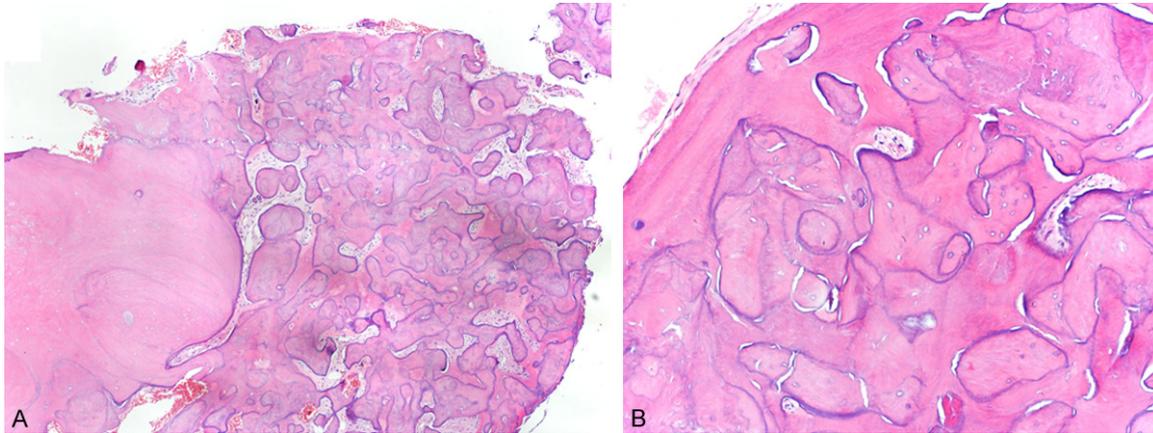


Figure 4. Hematoxylin and eosin-stained sections of the bone. (A) The lesion was composed of woven and lamellar bone, along with SMCM and some fibrous connective tissue. (B) No obvious osteoblastic and osteogenic reactions were observed. [Magnification, $\times 20$ (A); $\times 200$ (B)].

ing osteomyelitis, and Paget disease of bone, among others. As the histopathologic characteristics of the various types of COD do not vary greatly from each other, the patient's history, clinical manifestations, and radiographic examinations assist in differentiating one type from the others. Similarly, COD, OF, and FD are very similar in pathology, and are sometimes difficult to diagnose. OF occurs in 20- to 40-year-old women, and buccal and lingual swellings of the jaw are more apparent than in OF than in FLCOD. Furthermore, OF occurs mostly in the posterior part of the mandible and presents with a clear upper boundary when viewed on radiographs. FD, on the other hand, occurs mainly in adolescents; the clinical features include painless swelling often leading to facial asymmetry, occasionally accompanied by irregular café-au-lait spots, displacement of teeth, malocclusion, and in rare cases, root resorption. In addition, FD is distinguished by variations in the appearance of mineralized tissues, including the presence of woven bone. In the present study, panoramic radiographs did not reveal any similar lesions in the jaw bones of other member of the patient's family.

In the presence of infection, it is difficult to distinguish between FLCOD and chronic osseous sclerosing osteomyelitis. The latter is a primary infection and is accompanied by swelling and continuous pain. Small, scattered, dead bone particles that sometimes extend throughout the whole body of the mandible, including the ramus, are visible on X-ray. In contrast, FLCOD

occurs in the periapical, tooth-bearing region of the mandible, especially in the posterior area. Moreover, FLCOD is characterized by multiple lesions as is Paget's disease. However, the lesions are restricted to the jaws without involving the skull. Paget's disease, on the other hand, is most common in patients approximately 40 years of age and predominantly affects white males [17]. Serum alkaline phosphatase (ALP) is increased in patients with this bone disease; nearly 90% of patients with Paget's disease exhibited increased ALP levels in a study conducted in Japan [18]. However, the serum ALP level was within the normal range in the present case.

Conclusion

Herein, we present an extremely rare case of FOCOD and FLCOD occurring in the maxilla of a 68-year-old Chinese woman. FLCOD can grow out of the mandible similar to an exostosis. The progress of this lesion did not appear to be associated with the presence or absence of the related teeth. Further studies evaluating the etiology and associations between the various types of COD are warranted. Furthermore, it is also important to determine whether there is any relationship between FOCOD and FLCOD or whether they are two independent diseases similar to ossifying fibroma and fibrous dysplasia.

Disclosure of conflict of interest

None.

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