

Oral management of a patient with cemento-osseous dysplasia: a case report

Manejo oral de paciente com displasia cemento-óssea: relato de caso

Camila de Nazaré Alves de Oliveira **KATO**¹  0000-0001-9963-1707

Juliana Diogo de Almeida **SAMPAIO**¹  0000-0002-8419-5743

Tânia Mara Pimenta do **AMARAL**¹  0000-0001-8254-0497

Lucas Guimarães **ABREU**²  0000-0003-2258-8071

Cláudia Borges **BRASILEIRO**¹  0000-0001-6691-3943

Ricardo Alves **MESQUITA**¹  0000-0003-3207-4007

ABSTRACT

Florid cemento-osseous dysplasia is a subtype of fibrous-osseous lesion that commonly appears in the jaws of middle-aged black women. Clinically, the florid cemento-osseous dysplasia is asymptomatic. The radiographic aspect shows a radiolucent area with several masses of radiopaque structures, typically found in the toothbearing areas. Usually, the diagnosis is made during routine radiographic examination. The treatment is required when infection occurs due to exposure of the lesion in the oral cavity. In these severe cases, pain and facial deformity may be present. Commitment oral hygiene and routine dental visits for check-up are recommended for individuals affected. The objective of this study was to present a clinical case of a 24-year-old white woman with florid cemento-osseous dysplasia, who had been under surveillance for 13 years. Some changes in the radiographic pattern were observed as the orthodontic treatment was carried out in a private service. No tooth extraction was performed. This change is expected because florid cemento-osseous dysplasia has different radiographic aspects that correspond to the different developmental stages of the lesion. It is suggested, however, that the presence of florid cemento-osseous dysplasia did not preclude the success of the orthodontic treatment.

Indexing terms: Oral pathology. Orthodontics. Radiography.

RESUMO

A displasia cemento-óssea florida é um subtipo de lesão fibro-óssea que geralmente aparece nos ossos gnáticos de mulheres negras de meia idade. Clinicamente a displasia cemento-óssea florida é assintomática. Os aspectos radiográficos mostram uma área radiolúcida

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¹ Universidade Federal de Minas Gerais, Faculdade de Odontologia, Departamento de Clínica, Patologia e Cirurgia Odontológicas. Av. Antônio Carlos, 6627, Pampulha, 31270-901, Belo Horizonte, MG, Brasil. Correspondência para / Correspondence to: CNAO KATO. E-mail: <cnao20@yahoo.com.br>.

² Universidade Federal de Minas Gerais, Faculdade de Odontologia Departamento de Odontopediatria e Ortodontia. Belo Horizonte, MG, Brasil.

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com várias massas de estruturas radiopacas, tipicamente encontradas próximas às raízes dos dentes. O tratamento é requerido quando ocorre infecção devido à exposição da lesão na cavidade oral. Em casos mais severos, dor e deformidade podem estar presentes. Concomitante a higiene oral, visitas de rotina ao dentista são recomendadas ao indivíduo afetado. O objetivo deste estudo foi apresentar o caso clínico de uma mulher de 24 anos de idade, cor da pele branca com diagnóstico de displasia cemento-óssea florida com acompanhamento de 13 anos. Algumas alterações no padrão radiográfico foram observadas, como a mineralização progressiva da lesão. Tratamento ortodôntico foi realizado em um serviço privado. Não foi realizado exodontia. Esta alteração era esperada, pois a displasia cemento-óssea florida tem diferentes estágios de desenvolvimento. É sugerido, portanto, que a presença da displasia cemento-óssea florida não impediu o sucesso do tratamento ortodôntico.

Termos de indexação: Patologia bucal. Ortodontia. Radiografia.

INTRODUCTION

Fibro-osseous lesions are conditions that may affect the bones of the maxillofacial region. In general, the normal bone is replaced by cellular fibroblastic tissue containing calcified structures (osseous or cementum-like structures) [1-3]. According to the World Health Organization, the ossifying fibroma, familial gigantiform cementoma, fibrous dysplasia and cemento-osseous dysplasia have been included in the group of fibro-osseous lesions [4].

Cemento-osseous dysplasias (CODs) are reported as the most frequent fibro-osseous lesion in clinical practice. According to the place of manifestation and the extent, CODs may present three subtypes: periapical cemento-osseous dysplasia, focal cemento-osseous dysplasia and florid cemento-osseous dysplasia [4]. Florid cemento-osseous dysplasia (FCOD) is a more extensive form, occurring bilaterally in the mandible. The radiographic aspect of FCOD is characterized by multiple radiopaque areas with an aspect of lobulated masses [5,6]. If exposed to oral cavity, the acellular sclerotic bone can be infected, resulting in osteomyelitis frame [7,8].

All subtypes of COD have similar histopathological aspects and the diagnosis of FCOD is based on the clinical and radiographic features [1]. Due to the risk of infection, osteomyelitis and bone sequestrum formation, biopsy should be performed only in cases in which the radiographic and clinical findings are insufficient for a diagnosis [7,9]. Patients should be followed-up for observation of clinical and radiographic alterations [10,11]. Referral of patients with FCOD to orthodontics and other allied specialties of dentistry should be done carefully [12,13]. Taking into account the afore-mentioned statements, the aim of this study was to present a case of a 24-year-old woman with FCOD, who had been under surveillance for 13 years.

CASE REPORT

A 24-year-old white female was referred, in 2004, to the Department of Oral Surgery and Pathology at the Dental School of the Universidade Federal de Minas Gerais, for evaluation of a painless and hardened growth in the left posterior mandible. The medical and familiar history of the patient was non-contributory. The extra-oral examination revealed no significant finding. Intra-oral assessment showed the presence of a slight increase in the volume of the buccal alveolar bone in the area of the mandibular left pre-molars. Gingiva and oral mucosa showed no signs of inflammation. Filled and missing teeth were not observed.

The panoramic radiograph showed a radiopaque mass with a lobular pattern in a well-defined, round and radiolucent area around the apex of the mandibular left second pre-molar. A radiopaque area was also observed in the alveolar bone between the mandibular right first and second pre-molars and between the mandibular right second pre-molar and first molar (figure 1). The periapical radiograph of anterior region of the mandible showed bone changes close to the apex of the mandibular central incisors similar to those of the posterior region (figure 2). The mandibular incisors, the mandibular left and right pre-molars and the mandibular first molar responded positively to pulp vitality tests. Serum calcium, phosphorus, alkaline phosphatase, parathyroid hormone and vitamin D showed normal levels.

Due to bone expansion an incisional biopsy was carried out in the area of the mandibular left second pre-molar. This exam showed fragments of benign fibro-osseous tissue with the presence of calcified structures, represented by immature bone tissue with fibrous connective tissue (figure 3). Following the analysis of clinical, radiographic and histopathological features, the diagnosis of FCOD was confirmed.

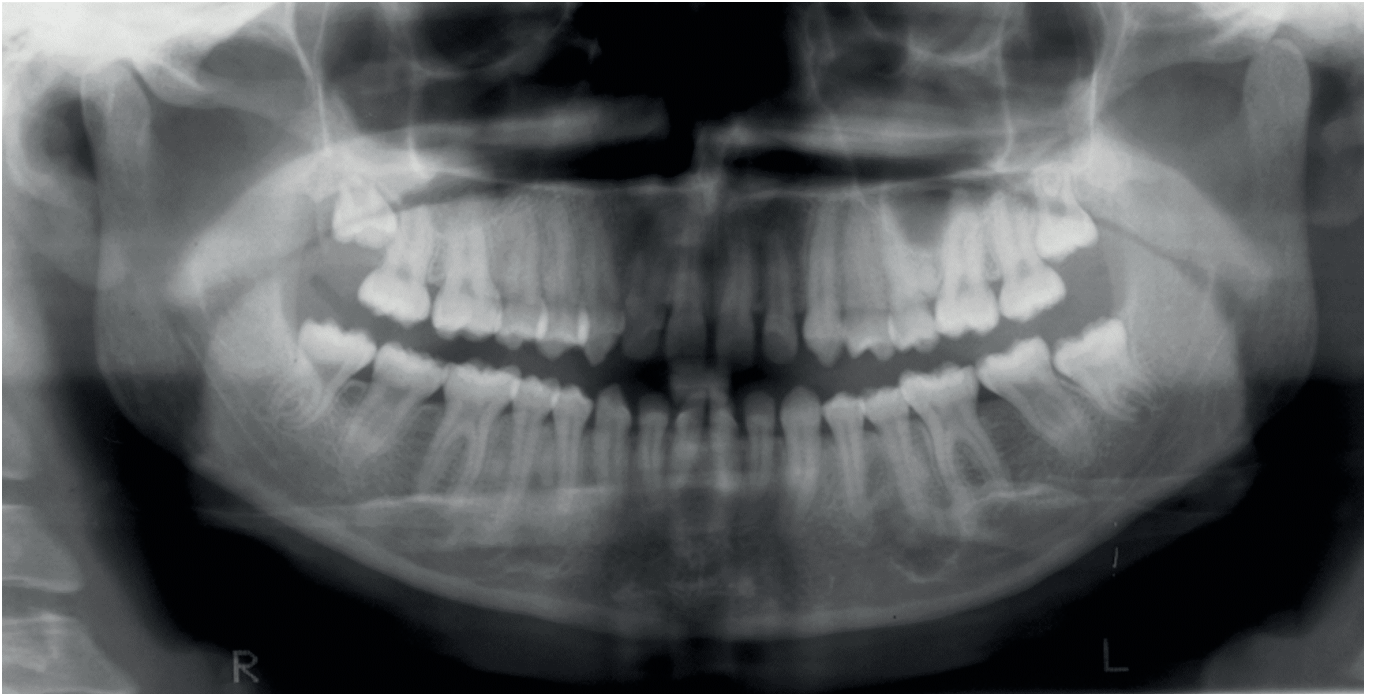


Figure 1. Panoramic radiograph in 2004 showing florid cemento-osseous dysplasia affecting mandible.

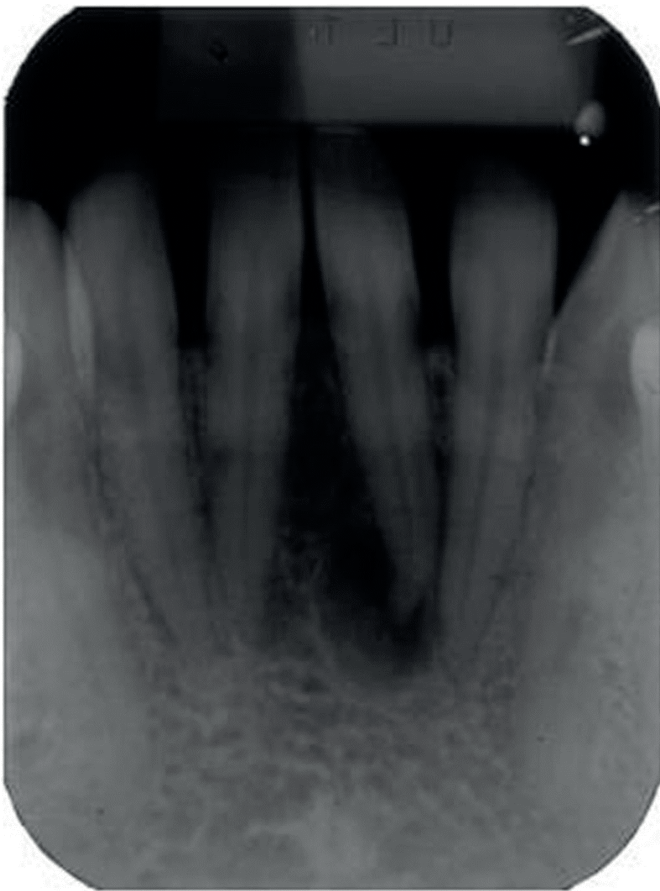


Figure 2. Periapical radiograph of the mandibular incisors area at early stage. Well-defined radiolucent area with small radiopacity inside.

The patient was placed under periodic surveillance to evaluate possible changes of the lesion (figure 4A). The clinical and radiographic exams, as well as pulp vitality tests of the teeth involved were performed every 2 years. In 2006, the patient manifested discomfort with the spacing between the teeth. Orthodontic treatment was carried out in a private service. No tooth extraction was indicated. The patient was advised to keep a strict regime of oral hygiene with adequate tooth-brushing and flossing as well as routine dental appointments for plaque control. Three years after orthodontic treatment onset, the panoramic radiograph showed a change in the radiographic pattern of the lesion in the left posterior mandible. A round radiopaque mass of calcified tissue was observed in contact with the root of the mandibular left pre-molar, surrounded by a radiolucent area. An increase of the lesion in the anterior mandible was also observed. On the day of orthodontic appliance debonding, panoramic radiograph demonstrated no change in the radiographic aspect in the area of the mandibular left second pre-molar. The lesion in the anterior mandible increased in size, extending from the mandibular right canine to the mandibular left canine. A radiopaque mass surrounded by radiolucent area was observed in this area (figure 4B).

In 2014, for more detailed examination, cone beam computed tomography (CBCT) was obtained (figures

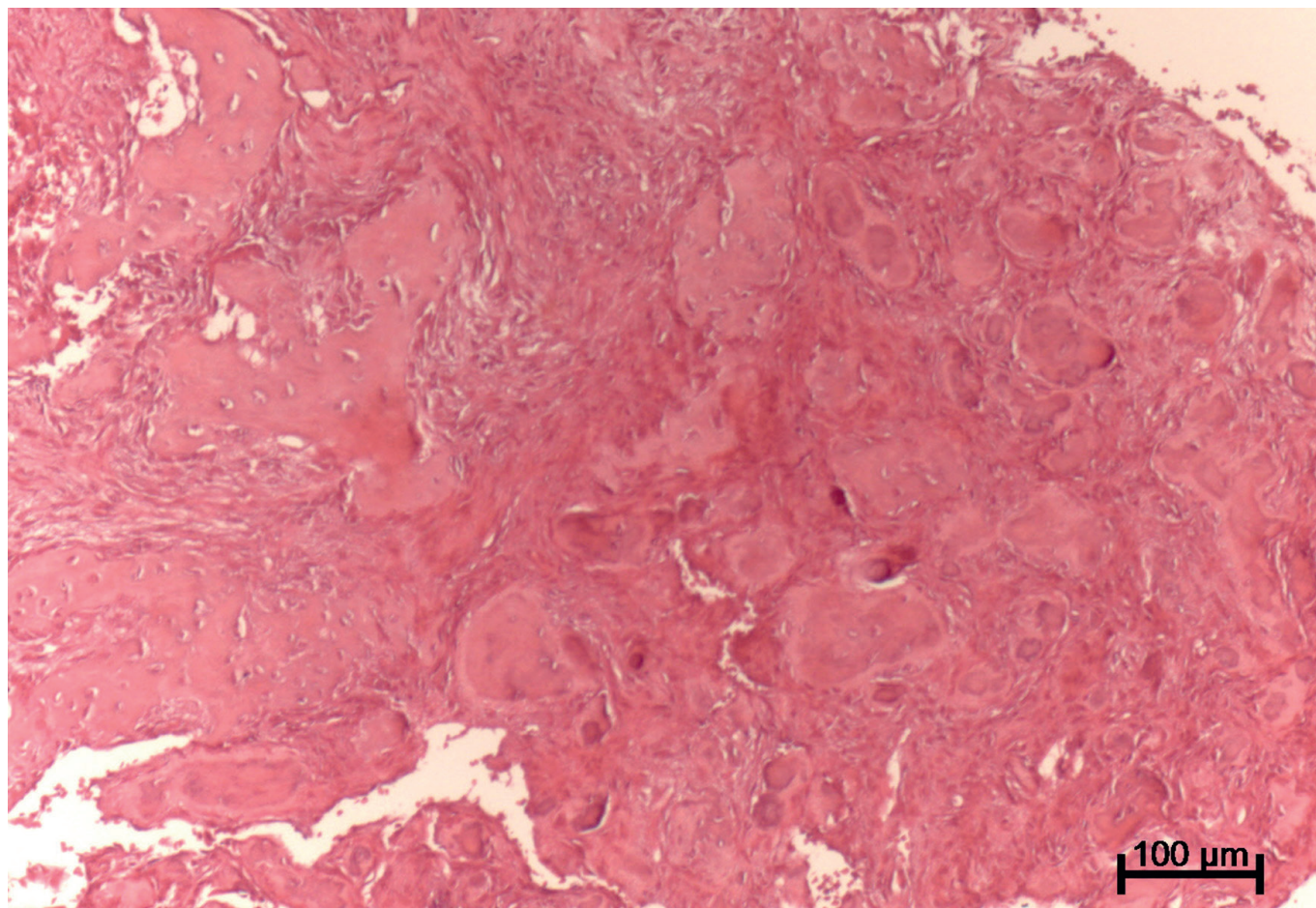


Figure 3. Histopathological aspect of the lesion.

5). Axial and coronal slice images revealed a hypodense and multifocal lesion with a hyperdense mass close to the mandibular left pre-molars. Expansion, without perforation, was also observed in the buccal and lingual cortical bone. Coronal slices showed an extensive high-density mass close to the mandibular left lateral incisor and the mandibular left canine with discontinuity or absence of the lamina dura of the periodontal ligament space.

During the surveillance, the patient has been encouraged to undergo routine dental appointments and to maintain strict oral hygiene to avoid inflammation that can affect the altered bone. In the last dental visit for check up, only panoramic radiograph was performed. An increased radiopacity was observed in the lesion (figure 6). Written informed consent of the patient was obtained for publication of the case report.

DISCUSSION

The etiology of the CODs remains unknown, but it may be associated with hormonal and genetic factors [5]. FCOD is most commonly observed among middle-aged black women [1,2,14]. The present case, however, describes a young female caucasian patient. Periapical and focal CODs affect restricted areas of jawbones and are usually asymptomatic. The diagnosis is made with routine radiographs. FCODs affect the mandible in 100% of the cases, while the maxilla is simultaneously involved in approximately 50% [15].

There are some cases of FCOD in which the patients report mild and intermittent pain in the affected area, especially after trauma or secondary infection [3,8,9,16]. In this case report, the patient has been followed up for a long period (thirteen years) and she has reported no

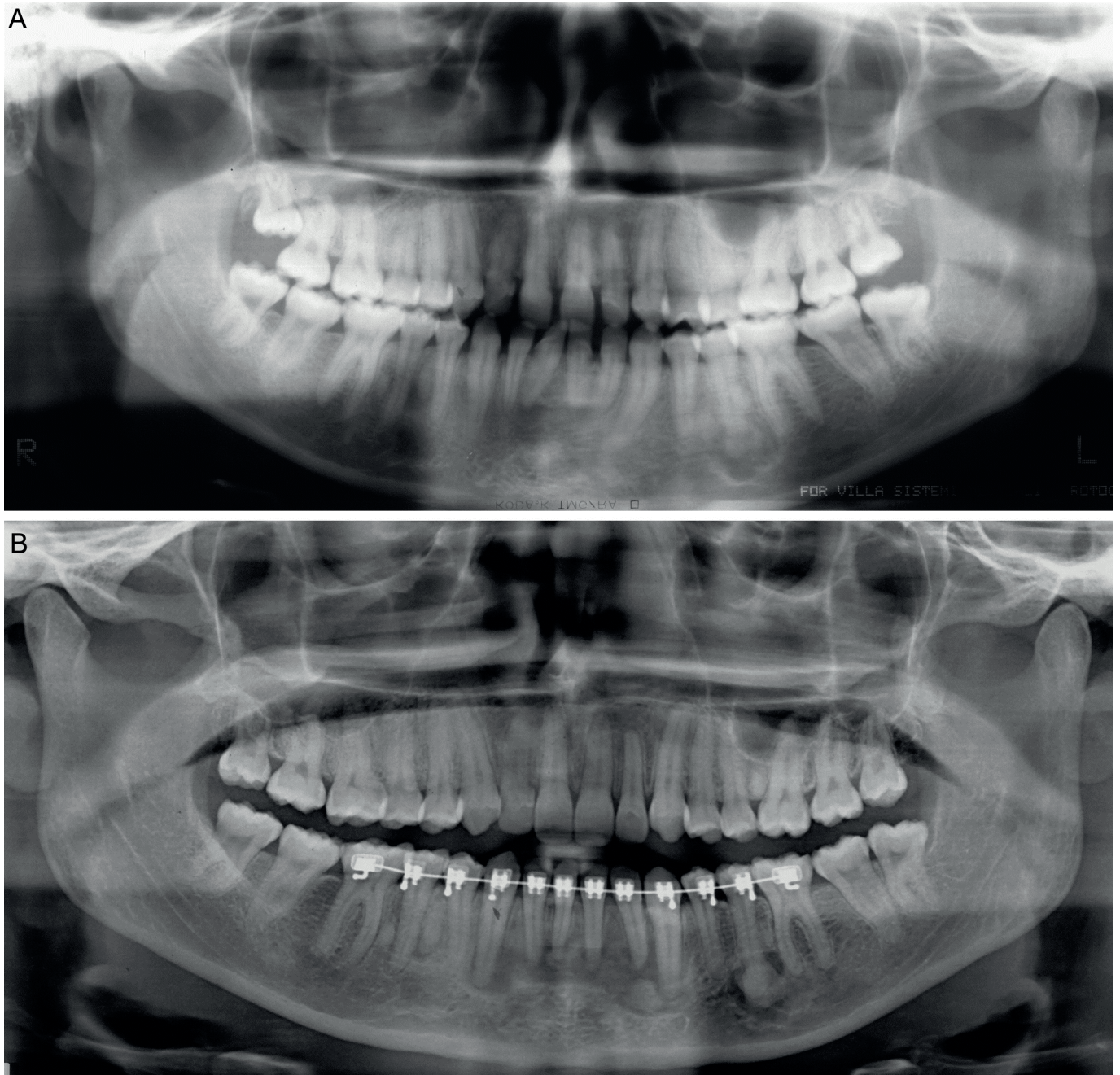


Figure 4. A) Panoramic radiograph in 2006. Patient under surveillance. B) Panoramic radiograph on the day of debonding. Increase of radiopaque areas within the lesion.

symptoms associated with the lesion. Only the swelling was observed. CODs are often limited to the alveolar process, leading to minimal expansion [6,15]. If cortical expansion with increase of the volume is present, a more in-depth investigation should be performed.

The diagnosis of fibro-osseous lesions is challenging because of the great similarity of features

among the lesions of the same group and other bone alterations as Paget's disease, chronic diffuse osteomyelitis [7,17] and inflammatory lesions [18]. Osteosarcomas and familial gigantiform cementoma should be also considered in the differential diagnosis. The combination of clinical, radiographic and histological data may contribute significantly to the final diagnosis of CODs.

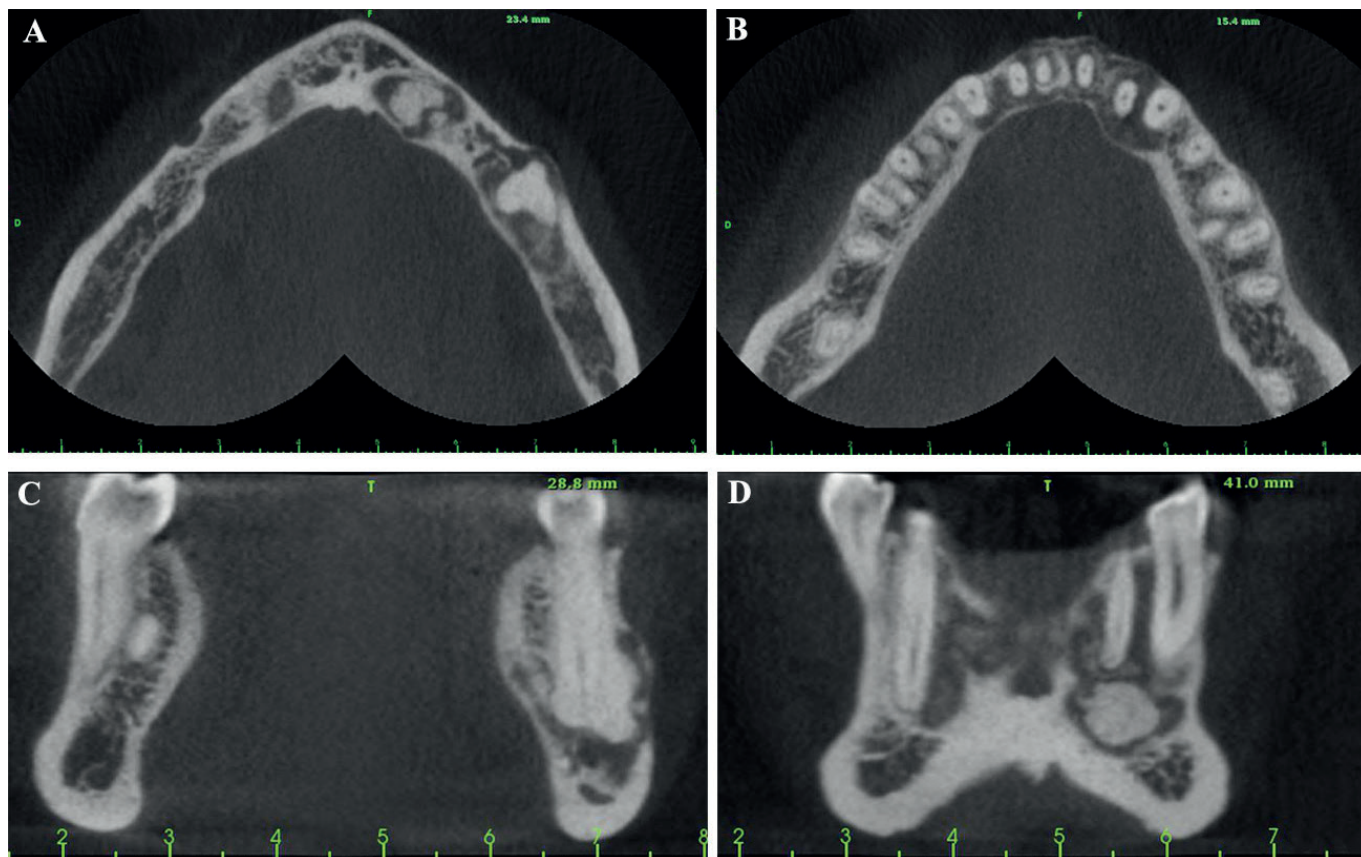


Figure 5. A) Axial slice of cone beam computed tomography taken in 2014 showing expansion of the buccal and lingual cortical. B) Superior axial slice showing expansion without perforation of the anterior buccal and lingual cortical bone. C) Coronal slice displaying discontinuity and absence of lamina dura in posterior region. D) Anterior coronal slice. Fibrous cement-osseous dysplasia causing discontinuity or absence of the lamina dura.



Figure 6. 13-year follow-up panoramic radiograph.

Careful observation of the characteristics of the lesion and a definitive diagnosis avoid unnecessary treatments [10,18,19]. In the present case, biopsy and histopathological exam was necessary to confirm the diagnosis of FCOD.

Management of asymptomatic patients requires periodic follow-up for clinical and radiographic evaluations. Another long follow-up of a patient with periapical COD has been reported elsewhere. This patient with a lesion affecting the anterior mandible had been followed-up for 12 years, but no intervention was carried out [10]. The author only stressed the importance of periodic control of the patient with oral prophylaxis and reinforced the relevance of good oral hygiene to prevent periodontal disease and tooth loss. The need for a careful plaque control has been emphasized in the literature [9,11,20,21].

Elective treatment, such as prosthodontic rehabilitation or orthodontic treatment among patients with CODs is little reported in the literature. In a clinical case of fibrous dysplasia, a normal response of dysplastic bone to orthodontic forces has been reported [22]. Successful cases of orthodontic treatment with no tooth extraction in young patients with FCOD have also been described [12,13]. A challenge in the orthodontic management of patients with COD occurs when tooth extraction is indicated. This procedure can result in the exposure of poorly vascularized sclerotic cemental masses in the oral cavity, which can lead to infection and consequent secondary osteomyelitis [1,2,8]. Pulp vitality tests should be carried out along with reinforcement of oral hygiene [12]. In the present case, no tooth extraction was performed during orthodontic treatment. The dentist stressed the need of rigorous oral hygiene control and performed pulp tests to confirm vitality.

During the orthodontic treatment, some changes in the radiographic pattern were observed. Progressive mineralization of the lesions close to the mandibular left second pre-molar and the incisors and canines were noticed. This change is expected because FCOD has different radiographic aspects that correspond to the different developmental stages of the lesion. In the osteolytic stage, a well-defined radiolucent area with loss of the lamina dura is observed. In the cementoblastic stage, radiopaque masses are seen in the radiolucent area due to mineralization (deposition of a cementum-like material). The final stage is characterized by a radiopaque mass in the lesion [5,7,11,15]. Thus, the radiographic changes observed from 2006 to 2017, during and after orthodontic

management, represent the usual developmental stages of the lesion.

Cone-beam computed tomography provides images from the axial, sagittal and frontal planes, and is indicated to evaluate the location and extent of the lesion [19,23,24]. Buccal and lingual expansion as well as a relationship with the anatomic structure was easily identified in the current case. Buccal expansion in the area of the mandibular left second pre-molar was observed. Lingual expansion of cortical bone in the mandibular left lateral incisor and canine area was also observed. Small expansions are observed in FCOD lesions, however expansions over 4 cm, with rapid growth without association with familial pattern have been recently reported [25].

CONCLUSION

The clinical case presented herein showed that changes in FCOD might take place over a period of time. Calcification and cortical expansion were observed in the imaging exams on the course of the follow-up. Surveillance of individuals with FCOD is highly encouraged in order to monitor the lesion's clinical behavior.

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Collaborators

The authors CNAO KATO and JDA SAMPAIO managed the patient clinically and drafted the manuscript. TMP AMARAL and CB BRASILEIRO drafted the manuscript and contributed to the acquisition, critical analysis and interpretation of the image. LG ABREU and RA MESQUITA were responsible for the conception of the article, drafting and editing of the manuscript. All authors read and approved the final version of the manuscript.

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