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CLINICAL SECTION

Orthodontic treatment and cemento-osseous dysplasia: a case report

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This report presents a case of florid cemento-osseous dysplasia (FCOD), affecting the mandibular teeth, in a 23-year-old Somalian female patient for whom orthodontic treatment was undertaken. The presenting features of the patient and the effect of orthodontic treatment on this case are discussed.

Key words: Orthodontic treatment, cemento-osseous dysplasia

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Introduction

Florid cemento-osseous dysplasia (FCOD) is a benign, fibro-osseous condition of bone.

Clinical findings

FCOD has a high incidence of occurrence in middle-aged African females.[1,2] A systematic review on FCOD reviewed 158 cases, of which there were 156 female cases, and found 59.6% (93 patients) were black, 37.2% (58 patients) were oriental and 3.2% (5 patients) were Caucasians and Indians.[1] The condition is most often painless and may only be detected when radiographs are taken for some other purpose. In severe cases focal expansion may occur, which may lead to pain[2] and facial deformities.

Radiographic findings

The radiographic appearance is characterized by multiple diffuse lesions that primarily surround the root apices of vital teeth. The lesions are often bilateral and have a symmetrical appearance. They can be found in the maxilla and the mandible[2,3] and are confined to the tooth-bearing regions.[4] Importantly, teeth are not a contributing factor in this condition. The lesions can vary from areas of radiolucency to radiopacity. Over time, the lesions can become increasingly radiopaque.[3,5] The shape of the lesions can vary from round to lobulated[4] and can range from being well to poorly defined.

Histological findings

Histologically, these lesions undergo change from normal vascular bone into woven bone in a matrix of fibrous connective tissue.[3,6]

Differential diagnosis

It is important to be aware of differential diagnoses as there are a number of other lesions that may have similar radiographic appearances.

Paget's disease of bone may have a cotton-wool appearance. It often affects the bone of the entire maxilla and mandible not just the alveolar bone and shows loss of lamina dura. It is often polyostotic, involving other bones such as the spine, femur, skull, pelvis and sternum.[2] In contrast to FCOD, it affects men more than women and is rare in those from African descent.

Chronic diffuse sclerosing osteomyelitis typically involves the mandibular body, angle and the ramus. This condition is usually associated with pain, swelling, fever and lymphadenopathy. The lesions demonstrate increased density and the borders are poorly defined. It can be distinguished from FCOD as it tends to extend beyond the tooth-bearing areas. Although women are

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more affected, black women are not particularly susceptible.[4]

Fibrous dysplasia is generally painless and lesions tend to stop growing after puberty is reached. The condition leads to superior displacement of the mandibular canal and various patterns of trabeculation have been described including 'orange peel' and 'ground glass'. It can be distinguished from FCOD due to the smaller size and peculiar shape of the lesions. In addition, the borders tend to be diffuse, blending in with the surrounding unaffected bone.

Osteosarcoma can often be differentiated radiographically by its 'sun-ray' appearance found in newly deposited periosteal bone. The lesions are more destructive extending into soft tissues, destructive of lamina dura and widening of the periodontal ligament spaces.

Periapical cemental dysplasia is a fibro-osseous lesion, considered a separate but related condition confined to the anterior mandible, within the space between the canines.[2]

Diagnosis

A radiology opinion is valuable in assisting in the diagnosis as diagnosis is largely from the clinical and radiographic features. Histology is rarely required as biopsy may precipitate infection which can be difficult to control without surgical intervention.

Management

The current methods of treating FCOD largely depend on the presence of symptoms. Where the patient is asymptomatic no intervention is needed, although it is advisable for the patient to undergo clinical and radiographic follow-up for several years to ensure that the lesions do not change in behaviour. Oral prophylaxis is important, with the emphasis on the control of periodontal disease and the prevention of tooth loss. The symptomatic patient is more difficult to manage. Symptoms may begin by exposure of the sclerotic masses into the oral cavity secondary to alveolar ridge atrophy or following dental extractions. This can lead to chronic inflammation due to the impaired blood circulation and the development of chronic osteomyelitis. Treatment usually involves surgical removal of the calcified fragments together with antibiotics with normal trabecular bone formation occurring after six months.

To the authors' knowledge, there have been no reports of the effects of orthodontic treatment in patients affected by FCOD. The aims of this case report were to examine the effects of orthodontic treatment on disease exacerbation, root resorption, tooth vitality and whether bone responds normally to orthodontic forces.

Case history

The patient, a Somalian female aged 23 years was referred by her general dental practitioner to the orthodontic unit because of her concern about 'the protrusive upper front teeth'. The medical history revealed mild asthma.

Extra-oral assessment

The patient presented with a mild skeletal II pattern due to mandibular retrognathia with an increased lower anterior face height and an increased Frankfort–mandibular planes angle (FMPA). There was an incompetent lip pattern with a lower lip trap. The incisor show was normal at rest and during smiling (Figure 1a–c).

Intra-oral examination

All permanent teeth were present excluding the upper third molars, lower left third molar and the lower left canine (Figure 1d–h). Oral hygiene was good and there were fissure sealants in the lower molars. There was bucco-lingual expansion of the alveolus in the region of the lower right first and second molars (Figures 1h and 2a) and expansion of the alveolar ridge superiorly, distal to the lower left second molar. The expansion was firm on palpation.

The lower incisors were of average inclination and there was mild imbrication. The upper incisors were proclined and spaced. In occlusion the incisor relationship was Class II division 1 with a 12 mm overjet and a symmetrical anterior open bite extending from the upper right first premolar to the upper left first premolar, measuring 3 mm maximally. The lower dental centreline was displaced to the left reflecting the absence of the lower left canine. The buccal segment relationship was Andrews' Class I bilaterally. There was a localized posterior crossbite of the upper right molars with no detectable mandibular displacement on closure.

Special investigations

Radiographs. A panoramic radiograph (Figure 2b) revealed two well-defined round radiolucent areas associated with the lower right first, second and third molars. There was a lobular mixed radiolucent and radiopaque lesion with nodular radiopacities extending from the lower right first premolar to the lower left first premolar which ranged from well defined on the right side to poorly defined on the left side. There was a further round, well-defined radiolucency distal to the lower left second molar. Cephalometric analysis confirmed all the previously mentioned clinical features.

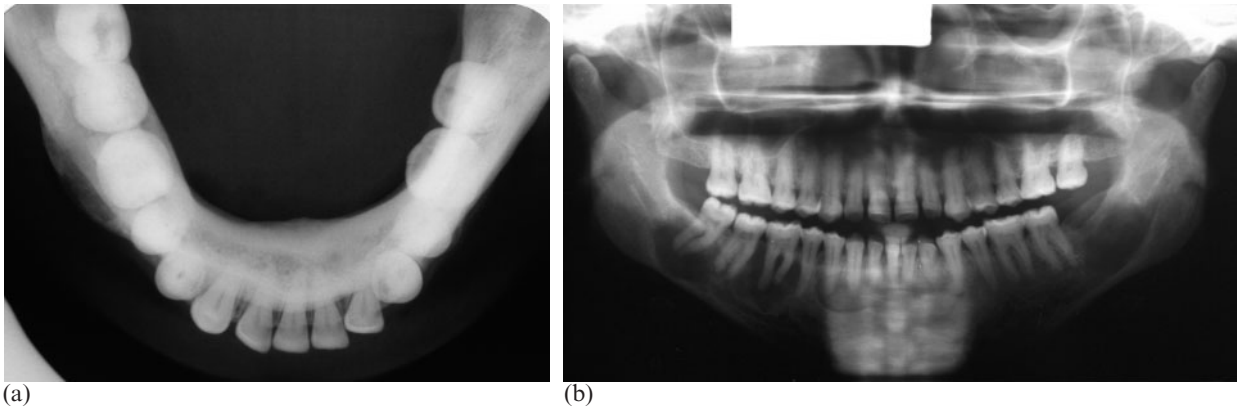
Vitality testing. All lower teeth responded positively to ethyl chloride and electro-pulse testing.

Figure 1 Pre-treatment photographs: (a–c) extra-oral views and (d–h) intra-oral views

Biopsy. The patient underwent incisional biopsies under a day stay general anaesthetic procedure of the lower right canine, lower right first premolar, lower right first molar and the lower left third molar regions.

Diagnosis

Histopathological examination showed features consistent with florid cemento-osseous dysplasia.



(a) (b)

Figure 2 (a) Pre-treatment lower occlusal radiograph. (b) Pre-treatment orthopantomograph

Treatment plan

- Liaise with an Oral and Maxillofacial Surgeon regarding timing of orthodontic treatment.
- Oral hygiene instruction.
- Non-extraction upper and lower fixed appliance therapy using the Tip-edge appliance.
- Long-term retention.

Rationale for treatment

Non-extraction treatment was chosen due to the well-aligned lower arch and spaced upper arch. The lower dental centre-line shift was accepted due to the potential associated morbidity of tooth extraction in FCOD. The patient was informed of the need for lifelong retention due to unfavourable soft tissue morphology.

Treatment progress

Following a period of review in the oral and maxillofacial surgery department, the patient was seen in the orthodontic unit to begin treatment. The importance of exemplary oral hygiene was stressed to the patient. The patient was treated using standard Tip-edge mechanics. During treatment the patient's oral hygiene deteriorated and she was immediately referred to the school of oral hygiene and therapy for instruction. A mid-treatment panoramic radiograph (Figure 3) was taken seven months into active treatment which revealed that the lesions associated with the right molars and the lower left third molar were largely unchanged. The lesion associated with the lower right first premolar to the lower left first premolar appeared to have increased in size and radiolucency with nodular radiopacities still present. New lesions were noted in the apical region of the lower left second premolar and first molar. The patient was debonded following eleven months of active treatment. All lower teeth responded positively to ethyl chloride and electro-pulse testing at debond. The end of treatment photographs are shown in Figure 4. The patient has been reviewed during her retention phase and an orthopantomogram was taken two years post-debond (Figure 5). The radiograph revealed that the lesions associated with the molars on the right side remain largely unchanged from the pre-treatment radiograph. The lesion associated with the lower right first premolar to the lower left first premolar appears to have reduced back to the size in the pre-treatment radiograph although there appears to be a reduction in the nodular radiopacities seen. The lesion associated with the lower left third molar appears to have become more radiopaque. The lesion associated with the lower left second premolar appears to have resolved, although the lesion associated with the lower left first molar has increased in size, remaining radiolucent.

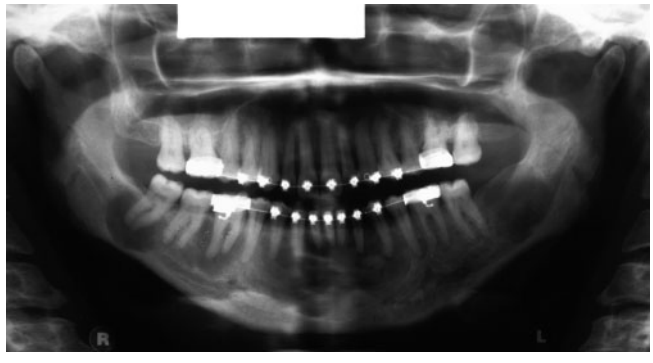


Figure 3 Mid-treatment orthopantomograph

Figure 4 Post-treatment photographs: (a–c) extra-oral views and (d–h) intra-oral views

Discussion

This case was diagnosed with FCOD following clinical, radiographic and histopathological examination. The literature reports the mean age of female patients presenting with FCOD to be between 47 and 49 years of age,[1] which is contrary to the age of the patient in this report, which is 23 years. It may be that as the lesions are often asymptomatic, presentation is noted later in life when individuals may be undergoing more extensive dental treatment. Familial FCOD has been reported[7] and affects younger individuals; however, there was no family history in this case. In addition, it does not have a predilection for African females and the rate of growth

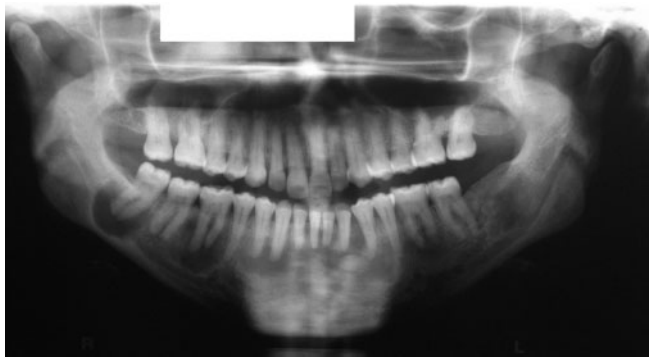


Figure 5 Two years post-debond orthopantomograph

is rapid. The importance of establishing tooth vitality in FCOD is paramount in excluding a dental origin and in reaching a correct diagnosis. Although it has been suggested that histological investigation is unnecessary in reaching a diagnosis of FCOD,[8] a biopsy was undertaken in this case due to the bony expansion noted in the mandibular molar regions. The patient was kept under review following diagnosis to ensure that the lesions had undergone no behavioural change.

Orthodontic treatment was initiated in the presence of good oral hygiene. Non-extraction treatment was undertaken in this case due to the well-aligned lower arch and the spaced upper arch. In orthodontic cases where the decision between an extraction and non-extraction approach is borderline, the latter may be more favourable as Waldron *et al.*[9] found that patients had poor socket healing and even sequestrum formation following extraction of teeth that were closely associated with large cemental masses. Some authors advocate not extracting in patients with FCOD.[9–11] This has implications for orthodontic cases that would be compromised without extraction. It may be that if extractions are inevitable that they should be as distant from the cemental masses as possible. If extractions are not possible, the feasibility of orthodontic treatment needs to be questioned.

Conclusions

This case shows that orthodontic treatment can be carried out in patients with FCOD in the presence of exemplary oral hygiene without fear of disease exacerbation, excessive root resorption or loss of tooth vitality. Extractions should be avoided if at all possible, especially if in close proximity to cemental lesions. In addition, teeth seem to respond normally to orthodontic forces.

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