Florid cemento osseous dysplasia: 15-years clinical and radiographic follow-up

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Abstract

Introduction
Florid cemento-osseous dysplasia is one of the terms that have been designated by the World Health Organization as cemento-osseous dysplasias of the jaws. The word 'florid' was introduced to describe the wide spread, extensive manifestations of the disease in the jaws. The condition is usually asymptomatic, however, in some cases symptoms of dull pain or signs of infection may be noted in the involved area. Florid cemento-osseous dysplasia (FCOD) has been described as a condition that typically affects middle-aged black women, and usually manifests as multiple radiopaque cementum-like masses distributed throughout the jaws. This paper reports a case of a 15 year clinical and radiographic follow up of florid cemento osseous dysplasia.

Case report
A case of asymptomatic uncomplicated florid cemento-osseous dysplasia occurring in a 59-year-old Jordanian female is reported, which can be considered rare regarding race distribution. The long-term follow-up (15 years) shows the different stages of maturation of the lesions and confirms that the condition is self-limited. The diagnosis was based on clinical and radiographic features only.

Conclusion
The diagnosis of FOCD mainly relies on radiographic and clinical findings. Vitality tests of related teeth are an essential addition to the diagnosis in order to avoid unnecessary endodontic therapy. Biopsy should be avoided in asymptomatic lesions as it may precipitate infection which might be difficult to control without surgical intervention.

Introduction
The WHO report 1992 describes Cemento- Osseous Dysplasias (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.1 The WHO report classify CODs on the basis of age, gender, location, histopathologic, radiographic and clinical characteristics.2

The term Florid Cemento-Osseous Dysplasia (FCOD) was first introduced by Melrose et al. in 1976 to describe a condition that probably represents an extensive manifestation of the reactive fibro-osseous process known as Cementoma.3 Different terms have been used to refer to cemento-osseous dysplasia COD of the jaws; these include periapical cemental dysplasia,4 florid osseous dysplasia,5 florid cemento-osseous dysplasia,6 focal cemento osseous dysplasia,7 and gigantiform cementomas.8

Florid cemento-osseous dysplasia FCOD is a rare condition presenting in the jaws of unknown aetiology.9 The lesion is more common in middle-aged black women,3,4 although it also may occur in Caucasians and Asians.10,15 In some cases, a familial trend can be observed.8,10,11

Clinically FCOD is a known self-limited disease.2 The process may be totally asymptomatic and discovered as a coincidental radiographic finding, it may cause facial deformity resulting from displacement of cortical bony plates.2,3 However symptoms such as dull pain or pus discharge may occur usually associated with exposure of sclerotic calcified masses in the oral cavity.3

Radiographically, the lesions appear as multiple sclerotic masses in the tooth-bearing regions which usually affect the mandible on both sides in asymmetrical manner, but all fourquadrants may be involved, it presents as multiple radiopaque lesions that fuse into lobulated sclerotic masses.12 They are often confined within the alveolar bone.3,13

Histological appearance of FCOD showed large sheets or fused globules of cemento-osseous tissue, which may be fused to the roots of one or more teeth or may lay separately.2 Lesions are composed of anastomosing bone trabeculae and layers of cementum like calcifications embedded in fibroblastic background.13

The purpose of this report is to present a case of FCOD which was diagnosed on the basis of clinical and radiographic features with 15 years follow up.

Case report
This report will present "according to our knowledge" the first documented case of FCOD in Jordan with 15 years clinical and radiographical data follow up. A 59-year-old Jordanian female patient attended the periodontal clinic at King Hussein Medical Centre in 1997 for periodontal assessment. Medical history revealed hyperlipidaemia controlled by statin group anti-lipidemic drugs and no other systemic disorders were reported.

Dental history revealed previous periodontal, restorative and endodontic therapies performed by different dentists during the treatment period which reported to be around thirty years; the last fifteen years were followed by the author of the present report. None of her family reported to have similar conditions.

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Extra-oral examination was within normal limits. Intra-oral examination revealed normal oral mucosa, good oral hygiene with minimal gingival inflammation, 4-5 mm periodontal pockets were found proximal to the maxillary posterior teeth with the deepest on the distal aspect of the maxillary left first molar, tooth wear was noticed on the incisal surfaces of the anterior teeth, and multiple restorative treatments mainly in the maxillary teeth.

Orthopantomograph (OPT) was requested and revealed multiple bilateral radiolucent-radiopaque sclerotic masses apical to the majority of mandibular teeth with no evidence of root resorption, dental caries was noted at the distal surface of the left mandibular first molar, poor endodontic treatment of the maxillary left first molar, generalized mild to moderate horizontal bone loss (Figure 1).

The left mandibular first molar was vital despite the radiolucency around the roots. The maxillary left first molar was symptomless. The treatment plan at that time included non-surgical periodontal therapy and restorative treatment of the caries on the mandibular molar in addition to keep the patient on periodontal maintenance and radiographic follow-up of the mandibular lesions. Blood investigation requested include complete blood count and full chemistry, all results were within normal limits except for cholesterol which was slightly elevated.

The patient was informed and reassured about her condition and the possibility of familial pattern, OPT was requested for her son and daughter, but no similar findings were evident. The patient was followed up for the next 15 years.

Figure 2 shows the OPT in 2003 which revealed the same symmetrical bilateral sclerotic masses in the mandible but with increasing of the radio-opacity in comparison with the OPT in 1997. The lower first left molar appears to serve as a satisfactory amalgam filling. The symmetrical radiolucency was remarkably reduced as more mineralization occurs at whole mandibular associated lesions. The radio-opacity keeps increasing with time in comparison with earlier records as shown in figure 3 which represents OPT images in 2005. The mandibular left first molar and the maxillary left first premolars were endodontically treated as a result of recurrent caries and crown fracture involving the pulp. Minor radiolucency could be recognized around the roots while the high dense deposit within the bone is obvious at the mandibular associated lesions.

Extractions of the mandibular left third molar and the maxillary left first molar were done in 2009 and 2012 respectively under antibiotic coverage of Amoxicillin 500mg three times daily for 5 days for periodontal reasons and without any difficulty or complications.

The latest OPG in 2011 as revealed in figure 4 demonstrates similar features with an increased density in comparison to earlier stages of the...
disease. No root resorption could be identified, and all the lesions are above the inferior alveolar canal.

Computed Tomography was requested at the same time in 2011 which shows a slight buccal expansion of the molar region the left side of the mandible as appeared in the horizontal and vertical sections (Figure 4). The patient is still under annual observation for periodontal maintenance, and three-yearly radiographic assessments for the mandibular lesions.

Discussion
Clinical and radiographic features are diagnostic for FCOD, it is almost always asymptomatic and nonexpanding, usually discovered incidentally on routine dental radiographs. The lesions usually present bilaterally in symmetrical pattern in the jaws start as radiolucent area at early stages which gradually becomes mixed radiolucent-radio-opaque, and finally appear as sclerotic masses with a thin peripheral radiolucent rim, when the lesion reaches the end stage it stabilizes and eventually does not cause any complication. In the present case, the patient was asymptomatic, and the first OPT was taken for periodontal assessment, whereas the mandibular lesions were observed incidentally for the first time. The subsequent radiographic findings follow the maturation pattern of FCOD, and the lesions are confined within the alveolus at a level corresponding to the roots of the teeth, above the inferior alveolar canal.

Paget’s disease, chronic osteomyelitis, and familial adentomatosis Coli Gardner’s syndrome are conditions that should be considered in the differential diagnosis in order to confirm the diagnosis of FCOD. Paget’s disease can have cotton wool radiopacity appearance and hypercementosis of teeth similar to FOCD, however Paget’s disease affects multiple bones and when it occurs in the jaw, the pathology affects the entire mandible, in addition to the elevated alkaline phosphate levels in Paget’s disease. In the current report, the radiographic appearance did not involve the entire mandible and the alkaline phosphate level was normal, while no other bony pathology was reported.

Since this case was asymptomatic and confined to the alveolar bone, the diagnosis of chronic diffuse sclerosing osteomyelitis was excluded. This condition is a primary inflammatory condition involving the body of the mandible from the alveolus to the inferior border and may extend into the ramus, cyclic episodes of unilateral pain and swelling are features of the condition. Florid cemento-osseous dysplasia may have similarities with jaw bone changes in familial adentomatosis coli Gardner’s syndrome which is not only characterized by skeletal changes, but by skin tumours and dental anomalies. These symptoms were absent in the present case.

Florid cemento-osseous dysplasia is frequently associated to black women. The patient in this report can be considered as a rare case with regard to race distribution. Florid cemento-osseous dysplasia may be familial with an autosomal dominant inheritance pattern, but there are only a few examples in the literature in which the familial pattern has been confirmed. In the present case, none of her offspring have similar radiographic features, therefore, no familial aspects of the disease could be established.

The Management of florid cemento-osseous dysplasia for asymptomatic patients consist of regular recalls and observations to ensure that the process is confined within normal limits of the disease and to detect any possible changes, as the disease may persist for indefinite periods of time without causing any symptoms. However if associated symptoms of pain and pus discharge arise and do not respond to other conservative treatment measures, surgical intervention may be warranted. Although the disease is not life-threatening, it may cause functional and cosmetic problems. The patient in this report has been followed up for three years without any further complications and the radiographic findings have not changed significantly.

References

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treatment modalities surgical intervention may be indicated.\textsuperscript{13}

Biopsy should be discouraged especially when no symptoms exist, hence biopsy may increase the risk of infection or fracture of the jaw and it is not normally justified to surgically remove these lesions, as this often requires extensive surgery.\textsuperscript{21}

Extraction is not recommended due to the poor socket healing from the impaired blood circulation in the affected area of the bone.\textsuperscript{22}

The management of our case consisted of conservative treatment and regular recalls, extractions were done due to a periodontal reason and under antibiotic coverage to minimize the risk of infection. Observations of similar asymptomatic radiopaque lesions over a long period enable us to predict their behaviour and ensure there are no negative changes of the lesion except what appears to be normal progression from radiolucency to radiopacity. Our findings are in agreement with other studies that reported long term follow up of florid cases without treatment.\textsuperscript{23}

\textbf{Conclusion}

The diagnosis of FOCD mainly relies on radiographic and clinical findings. Vitality tests of related teeth are an essential addition to the diagnosis in order to avoid unnecessary endodontic therapy. Biopsy should be avoided in asymptomatic lesions as it may precipitate infection which might be difficult to control without surgical intervention.

\textbf{Consent}

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

\textbf{References}


