Florid cemento-osseous dysplasia associated with chronic suppurative osteomyelitis and multiple impacted tooth an incidental finding – A rare case report

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Abstract

Florid cemento-osseous dysplasia (FCOD) is a rare, benign, fibro-osseous, and multifocal dysplastic lesion of the jaw that consists of cellular fibrous connective tissue with bone and cementum-like tissue. The word ‘florid’ was introduced to describe the wide spread, extensive manifestations of the disease in the jaws. FCOD is most commonly found in middle-aged, black women with the prevalence of 5.5%. Reports from Indian population is even rarer, with only 5 cases in literature, Florid cemento-osseous dysplasia is generally asymptomatic, and is usually detected during radiological examination. In this report, we present a case of a 44-year-old female patient diagnosed with Chronic diffuse osteomyelitis of the mandible later, on radiographic examination revealed FCOD, secondarily infected with Chronic diffuse osteomyelitis and multiple impacted tooth.

Keywords: Chronic suppurative osteomyelitis, fibro-osseous lesions, florid cemento-osseous dysplasia, impacted teeth

Introduction

Florid cemento-osseous dysplasia (FCOD), also known as Florid osseous dysplasia, Chronic diffuse sclerosing osteomyelitis, Sclerosing cemental masses of the jaws, Diffuse cementosis, Multiple enostosis, Gigantiform cementoma, Familial dominant cemental dysplasia is a benign fibro-osseous condition that affects jaw bones of middle-aged women, affecting two or more quadrants of the jaw, described by Melrose et al. for the first time in 1976. COD is a non-neoplastic, radiolucent and/or radiopaque, non-encapsulated, cellular fibrotic lesion with calcified structures, affecting the tooth-bearing area and the cancellous part of the jaws. The etiology of FCOD is unknown, and there is no clear explanation for its gender and racial predilections. 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.

Radiographically, FCOD is characterized by multiple masses of mixed radiopaque structures. 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. Biopsy for histopathological examination may not be required to confirm the diagnosis due to their characteristic radiological features. On the contrary, such attempt may increase the risk of infection or fracture of the jaw and hence will adversely affect the patient’s health.

As it is an incidental radiologic finding, which cannot be assessed in routine clinical diagnosis unless it is secondarily infected.

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Detection of such cases is of utmost importance to extend a hand of extensive primary care to any individual, which is the foundation of an effective diagnosis and treatment plan in order to achieve a better oral health outcome.

**Case History**

A 44-year-old female reported to the outpatient department of Oral Medicine and Radiology, with the chief complaint of a draining lesion from her chin region from the past four months.

Patient reported that she first experienced pain along with swelling in her lower right back tooth and gum region 1 year ago, for which she consulted a private dental practitioner where extraction of her lower right back tooth was done which was carious followed by course of antibiotics and analgesic which regressed the symptoms.

From the past 4 months, patient developed pain and swelling in the same area along with discharge from her chin region, pain was sudden in onset, dull aching, throbbing type, intermittent in nature of moderate intensity and localized to that region. Pain aggravates on mastication subsides on taking analgesic.

Her medical history was non-contributory and contained no history of previous trauma. Her past dental history reveals extraction of her lower right back tooth due to caries 1 year back. Fixed prosthesis on her upper and lower arch 2 year back.

Patient was well coordinated, moderately built and nourished, all vital signs were within normal limits.

On extraoral examination, an erythematous healing sinus present on the right submandibular region [Figure 1] measuring about 1 × 1 cm with retracted and crusted surrounding area and no discharge on applying digital pressure.

Intra-oral examination revealed sinus tract with purulent discharge at the level of buccal attached gingiva in relation to 45 surrounded by erythematous area [Figure 2]. On palpation there was buccal and lingual cortical plate expansion along with vestibular tenderness in relation to 44 45 region, edentulous space in relation to 46 47, severely attrited 45, fractured amalgam restoration 44, fixed prosthesis in relation to 16-26, 34-37, 48. Hence, correlating history and clinical examination, a provisional diagnosis of Chronic suppurative Osteomyelitis of the mandible was given.

An intra oral periapical radiograph taken in relation to 44, 45 [Figure 3], which revealed overhanging restoration in relation to 44, diffuse radiolucency at the periapical region of the root in relation to 45, loss of trabecular pattern in relation to 44, 45, multiple mixed radiopaque and radiolucent lesions in the periapical region in relation to 44,45 which was beyond the limit of IOPAR, hence a panoramic radiograph was taken [Figure 4]. It revealed multiple well defined radiopacity bilaterally on the maxilla and mandible in all the 4 quadrants limited to the tooth bearing area just above the inferior alveolar canal not extending beyond the 3rd molar region, multiple radiopaque lobulated structures of varying sizes, density similar to that of alveolar bone, with a thin radiolucent rim surrounding the radiopacity in relation to 35, 36, 37. Loss of trabecular pattern giving it a Moth-eaten appearance in relation to 44, 45. 13 23 18 were found to be impacted and missing tooth in relation to 12, 13, 14, 16, 23, 25, 26, 36, 37, 46, 47. Metal crown in respect to 17-27, 47, 34-37 region.

As the complete assessment of the lesion was difficult with a conventional radiograph, Cone Beam Computed Tomography was done for three-dimensional evaluation. Axial section of 1 mm thickness of CBCT showed a break in continuity of buccal and lingual cortical bone in relation to 44 45 region [Figure 5]. Coronal section showed multiple small diffuse radiopaque mass coalesce to form a larger radiopaque mass, with the greatest dimension of 15.72 × 10.55 mm in

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**Figure 1:** Extraoral (a). Shows extraoral sinus on the right submandibular region (b)

**Figure 2:** The intraoral periapical radiograph reveals overhanging restoration #44, mixed radiopaque and radiolucent lesion seen, with radiopaque mass at the center surrounded by radiolucent area giving it a moth-eaten appearance #44,45
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relation to 36 37 region in the mandible and 10.78 × 8.38 mm in relation to 26 27 region in the maxilla [Figure 6], mandibular canal was intact.

Patient was subjected to haematological investigation and alkaline phosphatase test where all parameters were within normal limits. As the patient was willing to receive immediate pain relief, under all aseptic precaution extraction of 44, 45 was done along with Incisional biopsy under LA and sutures were placed [Figure 7]. The specimen was sent for histopathological examination which revealed features of osseous dysplasia.

Hence, correlating the clinical, radiographic and histopathological features florid cemento-osseous dysplasia with chronic diffuse osteomyelitis of the mandible was formulated as the final diagnosis. At one-week follow-up, the patient showed complete resolution of the fistulous tract with a completely healed socket intraorally [Figure 8], patient is under regular clinico-radiological follow-up.

Discussion

Osseous dysplasias are a group of nonneoplastic fibro osseous lesions characterized by an alteration of bone structure and its replacement by fibroblasts, collagen fibers and mineralized material. A systematic review on FCOD showed only three Indian patients (<2%) identified from the whole series that combined most of the cases reported around the world.[7] Macdonald-Jankowski has conducted a study, who compared the prevalence of FCOD across the race. Of the 156 evaluated women who had the lesion 59.6% black women, 37.2% are oriental woman, 3.2% are Indian or Caucasian.[8,9] In the present case, patient was a 44-year-old female.

In the first World Health Organisation (WHO) classification of odontogenic tumours in 1971, four lesions referred to as “cementomas” were grouped together under the heading of “neoplasms and other tumours related to the odontogenic apparatus” (Pindborg et al., 1971).[10] Fibrous dysplasia and ossifying fibroma were placed in the category of “neoplasms and other tumours related to bone” (Pindborg et al., 1971) [Table 1]. Recently, in the 2017 publication the WHO classified COD under the category of “fibro-osseous lesions” (El-Naggar et al., 2017)[11] [Table 2].

Figure 3: Intra oral view shows swelling in buccal aspect of alveolus #44, 45, 46 region, with sinus tract at the level of buccal attached gingiva #45

Figure 5: CBCT axial section shows disruption of the buccal and lingual cortex in the right premolar region

Figure 6: (a) CBCT coronal section shows well defined radiopacity with greatest dimension of 15.72 × 10.55 mm #36.37 region. (b) CBCT coronal section shows well defined radiopacity with greatest dimension of 10.78 × 8.38 mm # 26.27 region

Figure 4: The panoramic radiograph reveals mixed radiopaque radiolucent lesion with ill-defined margins, presence of irregular central radiopacity and peripheral radiolucency giving a moth-eaten appearance in the right premolar region. Multiple radiopaque lesions with radiolucent rim surrounding it seen in maxilla and mandible
According to a study done by M. Benaessa, which suggested COD mainly affected the mandible (62.4%), followed by involvement of both the maxilla and the mandible (24.5%), and maxilla (13.1%). Of the 143 patients with known COD subtypes, florid COD predominated (65%), 33% of all cases of chronic suppurative osteomyelitis in this study were seen in patients with COD. Their occurrence in Indian population is rare and very few cases (less than 2%) have been documented in literature.

FCOD appears bilaterally, mostly in the mandible and often presents symmetrically. The term “florid” related to cemento-osseous dysplasia was introduced to describe the extensive manifestations of the disease in multiple quadrants of tooth-bearing area of the jaws. FCOD is usually asymptomatic and diagnosed accidentally at routine dental radiographic examination. FCOD can reach large dimensions and at times expose across alveolar mucosa. This exposed sclerotic bone is susceptible to infection progressing to a chronic osteomyelitis.

Chronic suppurative osteomyelitis exists when the portion of cancellous trabecula die and forms sequestra due to occlusion of regional blood vessels because of the pus accumulation spreading through the Harvesian and Volkman's canal which spreads along the medullary spaces breaches the periosteum, forms an extra oral sinus called cloacea.

FCOD tends to have an abated vascularization, which paves the path for the infection of these lesions. Which was also seen in our case.

Biopsy is not indicated as FCOD lesions are susceptible to infection leading to osteomyelitis due to avascular nature of altered tissues. Clinical and radiological features are most important to diagnose FCOD.

FCOD lesions should be differentiated from other similar appearing sclerotic lesions on conventional radiographs. Paget's disease manifests as cotton-wool appearance involving multiple bones such as maxilla, mandible, skull, pelvis and produces elevated alkaline phosphate levels whereas FCOD is centred above the inferior alveolar canal in mandible and serum alkaline phosphatase level is within normal limit.

There is no need for any treatment in an asymptomatic patient. Antibiotics are generally not effective in FCOD due to poor tissue diffusion, but regular follow up is mandatory due to the susceptibility to infection and fracture of the jaws, Revaluation with panoramic radiographs should be done in every 2 or 3 years in an asymptomatic patient.

In symptomatic cases, administration of antibiotics is indicated, but sometimes it may not respond to antibiotics due to the avascular nature of the lesion, requiring surgical debridement and enucleation.
Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflict of interest
There is no conflict of interest.

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