

Co-Occurrence of Florid Cemento-Osseous Dysplasia and Simple Bone Cyst: a Case Report

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ABSTRACT

Background: The purpose of this report is to present a rare case of co-occurrence of florid cemento-osseous dysplasia with simple bone cyst in a middle aged Asian woman. Most of the reported cases are isolated cases of simple bone cyst or florid cemento-osseous dysplasia, but co-occurrence of these two entities is extremely rare.

Methods: The authors report a 41 year old female patient with co-occurrence of mandibular florid cemento-osseous dysplasia with simple bone cyst. A thorough clinical and radiological examination was carried out.

Results: It was diagnosed mandibular cyst with possible co-occurrence of florid cemento-osseous dysplasia. Surgical exploration of the multilocular lesion was applied. Since, the patient was symptomatic at the time of presentation utmost caution was taken during the surgical procedure as florid cemento-osseous dysplasia is associated with hypo-vascularity of the affected bone. Based on histopathological, as well as supporting clinico-radiological findings a confirmative diagnosis of florid cemento-osseous dysplasia co-occurring with simple bone cyst was made. Patient was followed-up for a period of six months and was reported to be asymptomatic.

Conclusions: Timely diagnosis and well planned treatment is important to obtain a good prognosis when a rare co-occurrence of two or more bone lesions affects the jaws.

Keywords: florid cemento-osseous dysplasia; mandibular diseases; mandibular neoplasms; surgery, oral.

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INTRODUCTION

Florid cemento-osseous dysplasia (FCOD) refers to a group of fibro-osseous lesions, which are exuberant, multiquadrant and arise from the tooth-bearing area of the jaws [1]. FCOD is a benign jaw lesion and is discovered most frequently in the mandible of middle-aged black female [2]. It usually presents a multiple radiopaque bone/cementum-like masses distributed throughout the jaw [3].

FCOD lesions have a striking tendency towards bilateral, often quite symmetrical, location, and it is not unusual to find extensive lesions in all 4 posterior (molar-premolar region) quadrants of the jaw [4]. The word “florid” was introduced to describe the wide spread, extensive manifestations of the disease in multiple quadrants of the jaws [5]. The various synonyms used are multiple enostoses, multiple cemento-ossifying fibromas, multiple periapical osteofibromatosis, florid cemento-osseous dysplasia and gigantiform cementom [2]. In 2005, World Health Organization subdivided Cemento-Osseous Dysplasias (CODs) into periapical, florid and other CODs [6]. The definite diagnosis of these 3 diseases cannot be reached by clinical ground, but only by histopathologic examination [2]. Although, the disease may be totally asymptomatic, some patients present with pain, swelling, purulent discharge and sequestrum formation [7]. Rare reports of its association with simple bone cysts (SBC) are present usually when the cases reported are symptomatic. Melrose and co-workers [8] were the first to observe this association in their series of 34 cases, where 14 patients had concurrent, biopsy-proven simple bone cyst. FCOD is not associated with any other extragnathic abnormalities and there are no abnormalities in blood chemistry of patients [5].

Most benign fibro-osseous lesions of jaws are asymptomatic and slowly progressing [9]. Those benign fibro-osseous lesions that present as an atypical radiographic appearance require a detailed clinical, radiographic and laboratory workup to arrive at a diagnosis [10]. For the asymptomatic patient, the best management consists of regular recall examinations with prophylaxis and reinforcement of good home hygiene care to control periodontal disease and prevent tooth loss [11].

The case presented here highlights a rare combination of FCOD co-existing with a multilocular radiolucency. Computed tomography (CT), because of its ability to give three-dimensional axial, sagittal, and frontal views, is useful in the evaluation of these lesions. This paper presents the case of a patient, who was diagnosed with FCOD on the basis of

clinical and radiographic findings. SBC was diagnosed on the basis of surgical and histopathological findings. We hereby report one rare case of co-occurrence of florid cemento-osseous dysplasia with simple bone cyst presenting with symptoms of pain and swelling in the jaw.

CASE REPORT

41 year old female patient reported to the Department of Oral and Maxillofacial Radiology, AB Shetty Memorial Institute of Dental Sciences, Nitte University, Deralakatte, Mangalore, Karnataka, India, with a complaint of pain in the right posterior region of the mandible since 4 days. The pain radiated to the right side of the face, and was continuous in nature. Patient gave a history of extraction of a tooth in the same region 2 years ago. Tooth was extracted due to decay, after which timely healing did not occur and was associated with mild intermittent pain. Patient also complained of an enlargement of the jaw in the same site. No associated systemic symptoms were reported by the patient. Patient’s medical history was non-contributory. Extraoral examination revealed single diffuse smooth swelling on the right side of the jaw, approximately 1 x 1 cm in size, with normal overlying skin (Figure 1). The swelling was hard in consistency and tender on palpation. Right submandibular lymph nodes were tender on palpation. Intraoral examination showed an unhealed extraction socket seen in relation to #48. Surface was covered by slough and debris.



Figure 1. Clinical photograph of the patient showing swelling on the right side of the cheek (white arrow).



Figure 2. Intraoral photograph showing unhealed extraction socket (encircled in black).

Surrounding mucosa appeared to be normal in colour. No discharge was noticed (Figure 2).

Palpation revealed swelling with bony hard consistency and soft tender. Buccolingual cortical expansion was experienced. The area surrounding the socket was tender. Normal response was seen in all the teeth, when electric pulp testing was carried out.

An intraoral periapical view of the area revealed a well defined radiolucency. Superiorly, it extended 0.5 cm from the crest of the edentulous alveolar ridge and presented with scalloped border, with incomplete septa running into the radiolucency (Figure 3). The inferior and anterior extent of the radiolucency were not clear, hence an OPG was suggested.

Orthopantomography revealed well defined radiolucency in the body of mandible on the right side, measuring approximately 3 x 4 cm. It was present in the region of teeth #46, #47 and #48 involving the periapical area of #45 anteriorly, up to the anterior border of the ramus posteriorly. The radiolucent area extended superiorly up to 0.5 cm from the edentulous alveolar crest and 0.5 cm above the lower border of the mandible. The course of the mandibular canal could not be traced. Multiple mixed radio-opacities were present around the periapical region of all mandibular teeth above the level of the mandibular canal. The radiolucency was also observed at the periapices of teeth #18, #12, #26 and #28 in the maxilla suggestive of a fibro-osseous lesion. The radio opacities presented with a radiolucent halo around

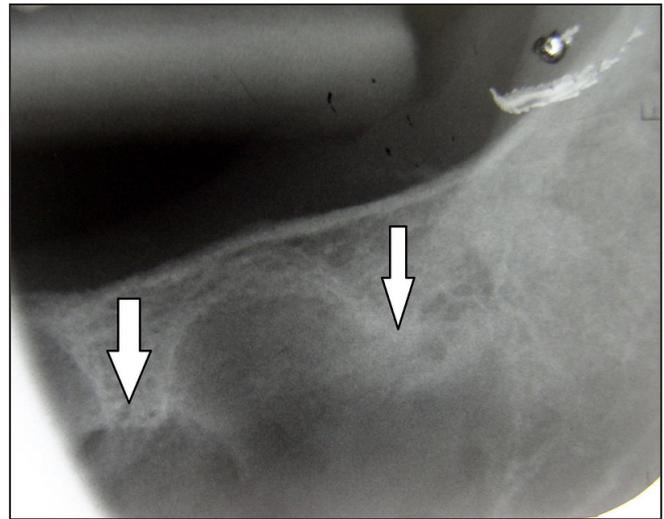


Figure 3. Intraoral periapical radiograph showing well defined radiolucency with scalloped border (white arrows).

them (Figure 4). Mandibular occlusal view of the same region revealed expansion of the buccal and lingual cortical plates with presence of multiple internal loculations (Figure 5). Serum work-up revealed no abnormalities.

Based on these radiological findings, lists of differential diagnoses for the radiolucency, as well as radio-opaque areas were made. For the multilocular radiolucency ameloblastoma, odontogenic keratocyst, simple bone cysts were enlisted and for the diffuse radioopaque lesion, diffuse sclerosing osteomyelitis, florid cemento-osseous dysplasia, Paget's disease were enlisted as differential diagnoses.

To determine the boundaries of the lesion preoperatively, a multidetector CT scan was made. It revealed, a well defined expansile osteolytic lesion measuring approximately 1.7 x 2.3 x 2.7 cm in size in the right body of the mandible at the level of the molar teeth.

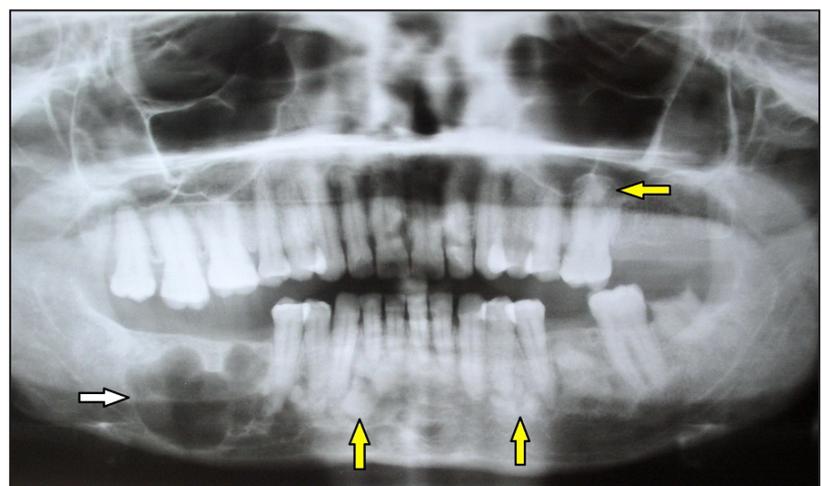


Figure 4. Orthopantomogram showing multilocular radiolucency on the right body of the mandible (white arrow) and multiple radio opacities in all the quadrants of both jaws (yellow arrows).

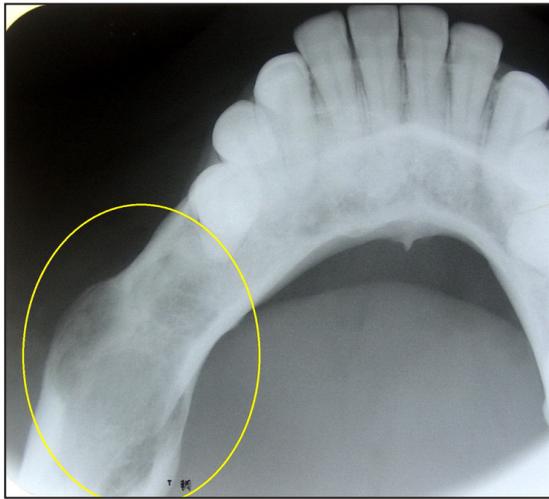


Figure 5. Mandibular occlusal radiograph showing cortical expansion and internal loculations (encircled in yellow).

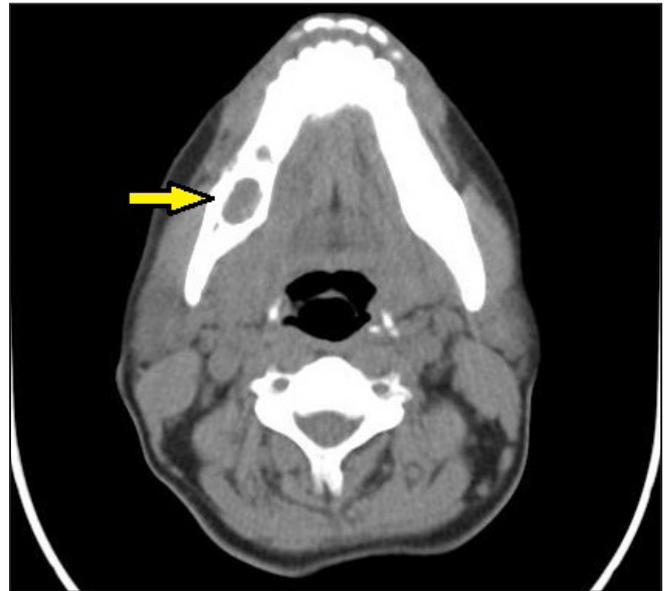


Figure 6. CT section showing a well defined expansile osteolytic lesion in the right body of the mandible (yellow arrow).

There is a narrow zone of transition (Figure 6). Multiple patchy well defined hyperdense lesions with hypodense rim were observed in the body of mandible located above the level of mandibular canal, between the inter-radicular and inter-dental areas of maxilla and mandible (Figure 7 and Figure 8). The CT scan findings were consistent with florid cemento-osseous dysplasia - stage III (mature stage).

Surgical exploration of the multilocular lesion showed connective tissue that was tightly adherent to the surrounding bone and was devoid of epithelium. The area was thoroughly cleaned with curette and the adjacent bony areas were sent for histopathological examination. Internal titanium plate fixation was done to the lower border of the mandible to avoid pathologic fracture due to thinning of the cortex (Figure 9).

Histological examination of the tissue lining the bony cavity revealed presence of fibrocellular connective tissue with osteoid areas as well as mineral deposition within them. The connective tissue shows spindle shaped fibroblasts and collagen fibres. Blood vessels and chronic inflammatory cells in the form of lymphocytes were additionally identified (Figure 10).

Histopathological examination of stained slides of periapical bone and buccal cortex in the region of teeth #45 and #46 shows immature bone with osteocytes. Numerous capillaries and resting lines are also seen (Figure 11).

Based on these histopathological, as well as supporting clinico-radiological findings a confirmative diagnosis of florid cemento-osseous dysplasia co-occurring with simple bone cyst was made.

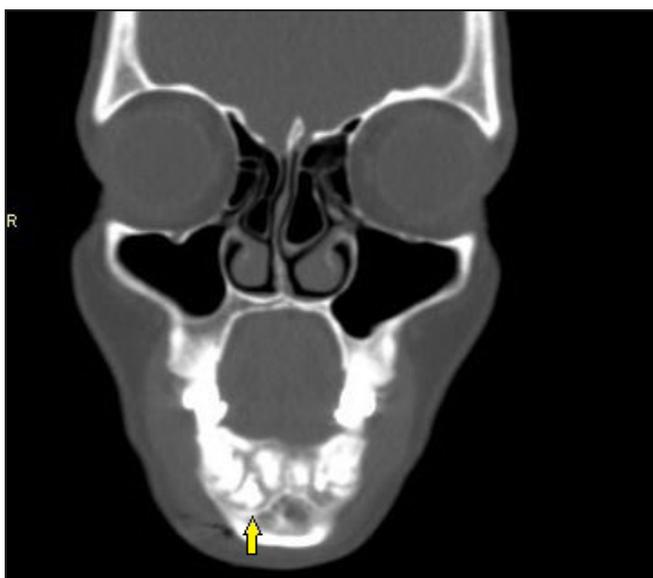


Figure 7. Coronal CT section showing multiple patchy well defined hyperdense lesions with hypodense rim were observed above the level of mandibular canal (yellow arrow).



Figure 8. Sagittal CT section showing multiple patchy well defined hyperdense lesions with hypodense rim (yellow arrows).

DISCUSSION

FCOD is the most common pathologic condition of the jaws that occurs as radiopacities in multiple quadrants of the tooth-bearing regions of the jaws [12]. This disorder is strictly localized to the tooth bearing areas and not associated with any other skeletal disease [13]. However, the etiopathogenesis is not clear. Waldron et al. [13] have proposed that reactive or dysplastic changes in the periodontal ligament might be a cause for the disease. This condition has also been classified by various authors as sclerosing osteomyelitis, sclerosing osteitis, sclerotic cemental masses, gigantiform cementoma, and various other terms [5].

FCOD is a benign fibro-osseous lesion in which mature bone is replaced with a woven bone in a matrix of fibrous connective tissue [14]. Similar histopathological features were noted in our case.

FCOD is a rare disease entity especially in the Indian population; only 2% cases have been reported among the Indians in the literature [13]. These lesions are most commonly seen in middle-aged black women, although it may also occur in Caucasians and Asians [3]. Our patient was a 41 year old woman of Asian origin.

Clinically, these lesions are often asymptomatic and may present as an incidental radiological findings [15]. FCOD occasionally is expansile, and patients may report experiencing pain [14]. Similar complaints were reported by our patient. Symptoms such as dull pain or drainage are almost always associated with exposure of sclerotic calcified masses in the oral cavity. No such features were seen in our case. This may occur as a result of progressive alveolar atrophy under a denture or after extraction of teeth in the affected area [16]. The patient presented herein underwent tooth extraction 2 years ago after which the healing did not occur. Occasionally patients may also present with complaint of intermittent, poorly localized pain in the affected bone, especially when a simple bone cyst has developed within the lesion. Extensive lesions often have an associated bone deformation [17]. Progressive increase in the bulk of the lesion was seen in case report presented by Miyake and Nagahata [18]. Similar history was presented by our patient. Waldron suggested that periapical cemento-osseous dysplasia and focal cemento-osseous dysplasia may also develop into florid cemento-osseous dysplasia [19,20].

A simple bone cyst can be found in association with benign fibro-osseous lesions such as cemento-osseous

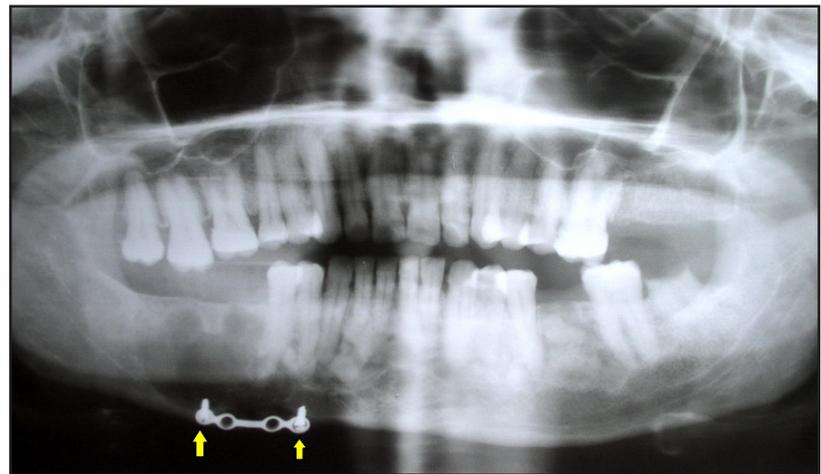


Figure 9. Postoperative orthopantamogram showing internal plate fixation (yellow arrows).

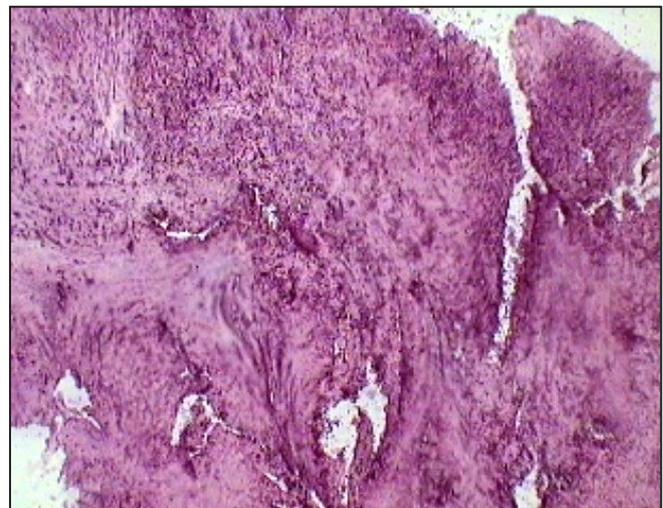


Figure 10. Hematoxylin and eosin stained histopathological section (original magnification x10) of periapical bone and buccal cortex in the region of teeth #45 and #6 showing immature bone with osteocytes.

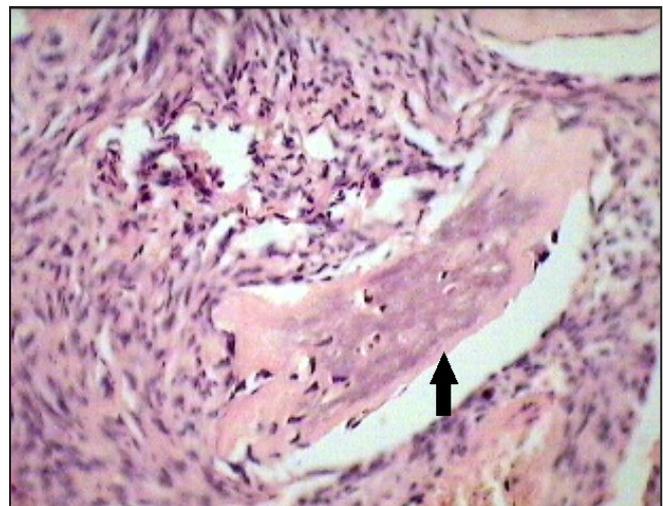


Figure 11. Hematoxylin and eosin stained histopathological section (original magnification x40) of cavity lining showing fibrocellular connective tissue with osteoid areas within them (black arrow).

dysplasia and fibrous dysplasia [15]. Some reports have described an association between solitary bone cysts and fibro-osseous lesions including fibrous dysplasia and cemento-osseous dysplasias [21]. FCOD with concomitant simple bone cysts is not common [22]. The pathogenesis of simple bone cysts is largely known. Venous obstruction and blockage of interstitial fluid drainage, in these areas of rapidly growing and remodelling cancellous bone, may lead to formation of the simple bone cysts [15].

In our case, the diagnosis of FCOD was made clinico-radiologically and histopathologically. An elective surgical procedure including biopsy was not performed, because of the risk of secondary infection. Secondary infection occurs in such lesions due to abundant cementum formation and poor vascularity [17].

Radiographically, FCOD is characterized by extensive sclerotic areas, often involving the posterior quadrants of the mandible and maxilla in a symmetric fashion [23]. FCOD is a diffuse form characterised by multiple periapical lesions involving one or both the jaws. It occurs around the root apices of vital tooth in middle-aged women with a predilection for mandibular incisors. In the early stage, it appears as a well defined radiolucent lesion, which gradually becomes totally radiopaque with a thin lucent rim in the mature stages [24].

Asymptomatic lesions do not require intervention because, complete resection of the lesion would be impractical as it usually occupies most of the mandible and maxilla [16]. When surgical intervention is indicated, a remodelling resection is recommended for aesthetic reasons [25]. Histopathologically, FCOD is composed of a proliferating fibrous connective tissue stroma containing foci of cementum along with the presence of osteoid or bone. More advanced lesions show an increase in mineralization. In FCOD, large sclerotic masses are formed that are hypocellular and extremely dense with small marrow spaces and few Haversian systems [21].

The simple bone cysts that occur without any association with cemento-osseous dysplasia tend to heal better after surgery than those associated with cemento-osseous dysplasia [26]. The histological features of simple bone cysts are mostly non-specific, and when a tissue is submitted from this lesion, a microscopic examination will reveal only a strip of fibrous connective tissue,

occasionally with an associated rim of bone [15].

Expansion of the buccal and lingual cortical plates is not observed in FCOD unless associated with cystic changes [27]. A similar finding supported our case.

As differential diagnosis we can include Paget's disease which would present radiologically with cotton-wool appearance and diffuse sclerosing osteomyelitis which can also be a complication associated with FCOD. Paget's disease is also characterized by deformities of multiple bones and produces biochemical serum changes, such as elevated alkaline phosphate levels [15]. No such biochemical alterations or other bone deformities were present in present case.

Regular follow-up during six month was maintained and patient reported to be asymptomatic after the surgery.

CONCLUSIONS

The management of florid cemento-osseous dysplasia may not be satisfactory, since the disease process may run for a very long time without any symptoms. When patients are asymptomatic, optimum oral hygiene has to be maintained to avoid tooth loss and periodontal disease. Elective intraoral procedures have to be avoided due to the associated risk of infection or subsequent osteomyelitis and fracture. When the patient is symptomatic secondary to a tooth pain, the tooth may be managed endodontically by avoiding extractions. Cases with secondary predisposed factor of infection are difficult and complicated to manage. Since our patient will require frequent scans during the follow-ups, imaging modalities with lesser radiation doses have to be employed. Surgery, when required should be carried out with a minimal invasion. The treatment options should be easily available and accessible, affordable to the masses without hampering the aesthetics and function of the patient.

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