Florid Cemento-Osseous Dysplasia-Report of Rare Case

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Abstract
Florid cemento-osseous dysplasia is a rare disease of jaws which is characterized by replacement of bone and connective tissue by cemento-osseous tissue. They are mostly found in middle-aged individuals. They are generally found in tooth-bearing region involving two or more quadrants. Radiographically, these lesions appear as multiple sclerotic masses confined within the alveolar bone. Herein we present a rare case of florid cemento-osseous dysplasia involving maxilla and mandible.

Keywords: Fibro-osseous lesions, Florid cemento-osseous dysplasia, florid osseous dysplasia

Case Report
A 41 year old female patient (Fig.1) reported with chief complaint of swelling in mandibular anterior region since 6 months. The swelling was progressively increasing in size causing mild facial asymmetry in mandibular anterior region. There was no significant past medical and dental history. The patient was normal built having normal gait and posture. Her vitals were within normal limit. She was well oriented to surroundings. There was no cervical lymphadenopathy, pallor and clubbing. On extra-oral examination there was mild asymmetry in mandibular anterior region (Fig.1). On intraoral examination a non-tender, firm to hard swelling of 3x3 cm noted which was extending from right mandibular canine to left mandibular canine. The swelling was extending in mandibular labial sulcus causing fullness of mandibular anterior region (Fig.2). Lingually the swelling was extending from right mandibular canine to left mandibular canine causing bicortical expansion but the severity of expansion is lesser in lingual side comparatively (Fig.3). The teeth from right mandibular canine to left mandibular canine were severely attrited involving mandibular premolars bilaterally also (Fig.3). The mucosa overlying the swelling was normal in colour and texture. Rest of structures were within normal limit. There was no associated pain and bleeding. The patient was advised for routine radiographic examination including IOPA, mandibular true occlusal radiograph and panoramic radiograph. The IOPA radiograph (Fig.4) showed multiple sclerotic masses (mix radiopaque radiolucent lesion) associated with roots of 31,32,33,34 and 41,42,43,44 confined within the alveolus. Ill defined radio opacity was surrounded by radiolucent rim. The lamina dura was well defined and intact. The mandibular true occlusal radiograph (Fig.5) showed bicortical expansile mix radiopaque radiolucent lesion with multiple discrete radio-opacity in relation to roots of 31,32,33,34 and 41,42,43,44. Panoramic radiograph (Fig.6) showed a well-defined mixed radiopaque radiolucent lesion surrounded by sclerotic margin extending from 35 to 45 region, 4x4cm (approx) in size and involving the anterior portion of mandible crossing the midline and confined within the alveolus. Multiple radio-opacity surrounded by radiolucent rim is noted in relation to 31,32,33,34 and 41,42,43,44. There was thinning of inferior cortex in region of lesion. Lamina dura of associated teeth were breached and was absent in some teeth. On the basis of routine radiographic investigations patient was advised for contrast enhanced CT. The CECT shows multiple expansile, osteolytic lesion in maxillary (Fig.7) and mandibular alveolar process (Fig.8) involving anterior part of body of mandible and both maxilla. The lesions are noted around the periapical area of maxillary and mandibular incisors and canines and involving right lower posterior teeth. Overlying cortical thinning with few areas of breach is noted with multiple foci of calcifications and patchy area of ground glassing. CT was useful to find out lesion in maxilla also which was clinically insignificant. The patient was advised for incisional biopsy. The routine blood examination was found to be normal and incisional biopsy has been done under local anesthesia. The histopathological micrograph (Fig.9) showed thick, confluent curvilinear trabeculae with little fibrotic stroma. This lesion showed formation of dense sclerotic calcified cementum-like masses. Periphery of the lesions showed globular or ovoid structures of cementoid appearance involved by thin fibrous tissue (H&E stain, ×200).
Fig. 1: Profile view of patient showing mild facial asymmetry in mandibular anterior region.

Fig. 2: Intra-oral view showing a swelling of 3x3 cm extending from right mandibular canine to left mandibular canine. The swelling was extending in mandibular labial sulcus causing fullness of mandibular anterior region.

Fig. 3: Intra-oral view showing lingual extension of the swelling from right mandibular canine to left mandibular canine causing bicortical expansion but the severity of expansion is lesser comparatively.

Fig. 4: IOPA radiograph showing multiple sclerotic masses (mix radiopaque radiolucent lesion) associated with roots of 31,32,33,34 and 41,42,43,44 confined within the alveolus.

Fig. 5: The mandibular true occlusal radiograph showing bicortical expansile mix radiopaque radiolucent lesion with multiple discrete radiopacity in relation to roots of 31,32,33,34 and 41,42,43,44.

Fig. 6: Panoramic radiograph showing a well defined mixed radiopaque radiolucent lesion surrounded by sclerotic margin extending from 35 to 45 region, 4x4 cm (approx.) in size and involving the anterior portion of mandible crossing the midline and confined within the alveolus. Multiple radio-opacity surrounded by radiolucent rim is noted in relation to 31,32,33,34 and 41,42,43,44.
Discussion

Florid cemento-osseous dysplasia is a rare disease which is characterized by replacement of bone and connective tissue by cemento-osseous tissue affecting the jaw bones. They are mostly found in middle-aged individuals characterized by radiopaque cementum-like masses involving multiquadrant of dentition. On the basis of extent and radiographic appearances the cement-osseous dysplasia is classified in 3 types, periapical type is typically surrounds the periapical region of teeth and are usually bilateral, florid type is characterized by sclerotic symmetrical masses and focal type is generally found as single lesion. The term florid was coined by Melrose et al. Melrose et al denoted the term florid to describe widespread and extensive nature of florid osseous dysplasia. However the term ‘florid cemento-osseous dysplasia’ is coined by Waldron due to close resemblance of the dense, sclerotic masses to cementum of tooth.

The term florid cemento-osseous dysplasia has been proposed in the second edition of International histological classification of odontogenic tumours to replace the term gigantiform cementoma given in the first edition’s, The WHO histological typing of odontogenic tumours: A commentary. The World Health Organization has classified cemento-osseous dysplasia on the basis of age, gender, location, histopathologic, radiographic and clinical characteristics. The florid cemento-osseous dysplasia is prevalent in African-American females in the 4th to 5th decade of life with mean age of occurrence 42 yrs of age. The etiopathogenesis of florid osseous dysplasia is still not clear. Some authors agrees on concept that the proliferation of the fibroblastic mesenchymal stem cells in the apical periodontal ligament which are considered as cement blastic precursor stem cells leads to formation of agglomerated mass while others agreed on the view that florid osseous dysplasia may arise from the remnants of the cementum left after tooth extraction. The FCOD can reach upto size of 1-2 cm and effects edentulous jaws and the tooth extraction sockets however it can be localized near teeth. FCOD can cause bony expansion and can be secondarily infected.

Previously the FCOD was reported under variety of pathologies like multiple cemento-ossifying fibroma, sclerosing osteomyelitis, multiple enostosis and gigantiform cementoma. Many authors have coined FCOD as paget’s disease of mandible and periapical cemental dysplasia. The focal COD arises at the previous extraction site or at the apices of the molar region, rarely exceeding 2 cm in diameter, mimicking the radiological presentation of the periapical osseous dysplasia. The FCOD and periapical osseous dysplasia involve a single area or multiple areas in a quadrant. The symmetrical presentation of pathology affecting more than one quadrant confirms the diagnosis. The FCOD are clinically asymptomatic and may be found as...
incidental radiological finding presenting as multiple radiopaque masses within peripheral radiolucent rim located in two or more quadrants usually in tooth-bearing areas. However sometimes associated symptoms such as dull pain or drainage is almost always associated with exposure of sclerotic calcified masses in the oral cavity. This may occur as the result of progressive alveolar atrophy under a denture or after extraction of teeth in the affected area. Broadly on the basis of clinical findings FCOD is divided in sporadic type and familial type. The sporadic cases have single site involvement however the familial form appears to be inherited as an autosomal dominant trait with variable phenotypic expression which is characterized by more expansile lesions and tends to occur in younger subjects. Radiographically these lesions appears radiolucent initially. With time the lesion evolves to a mixed radiolucent radiopaque stage before progressing to a completely radiopaque stage however there is absence of normal trabecular pattern of bone in the lesion. The lesions are typically found in the tooth bearing areas of the jaws. The radiographic appearance of FOCD is very characteristic and very helpful in establishing the diagnosis. CT is imaging modality of choice for differentiating FCOD from lesions exhibiting sclerotic appearance. The differential diagnosis of FCOD includes Paget’s disease, chronic diffuse osteomyelitis and Gardner’s syndrome, enostosis or exostosis and odontogenic tumors, especially cemento-ossifying fibroma.

Although, FOCD represents a spectrum of lesion rather than an entity therefore the histopathology will remain similar for all the types. The early lesion represents cellular fibrous tissue containing trabeculae of woven bone with cementum-like calcification. With maturation, the ratio of fibrous tissue to mineralized materials are decreased and trabeculae become more curvilinear structures that progress to the radiopaque stage in which the cementum-like tissue coalesce to form large basophilic calcification with resting and reversal line. Lesions are composed of anastomosing bone trabeculae and layers of cementum like calcifications embedded in fibroblastic background.

Management of FOCD involves clinical and radiographic follow-up. Until the definitive diagnosis is not obtained clinically and radiologically the endodontic therapy should not be considered. The lesions causing pain and cortical expansions surgery and the risks it entails might be necessary for adequate treatment. However recontouring is the treatment of choice where there is only cortical expansion and mucosal perforation due to the cement-ossesous lesions.

Conflicts of Interest: None
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References