Florid cemento-osseous dysplasia presenting as bilateral mandibular swellings – report of a rare case

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ABSTRACT

Florid cemento-osseous dysplasia, a benign non neoplastic fibro-osseous lesion more commonly occurs in middle aged Asian females. Most of the reported cases were asymptomatic. We report a case occurring in a middle aged Asian male with bilateral mandibular swelling, which is a rare finding alongwith radiographic and histopathological diagnostic challenges.

Keywords

Florid cemento-osseous dysplasia, fibro-osseous lesion, mandibular swelling, cone-beam computed tomography, case report

INTRODUCTION

Florid cemento-osseous dysplasia (FCOD), a member of fibro-osseous lesions is a benign non neoplastic lesion which shows replacement of normal bone with cellular fibrous tissue and immature bone maturing into cementum like material.[1] It is characterized by multifocal involvement affecting multiple quadrants of jaw bones.[2,3] It is asymptomatic initially and often discovered during radiographic investigations. Pain and swelling may appear if it gets secondarily infected requiring clinical intervention.[1] It is more commonly seen in middle aged African and Asian women.[2] This case report describes rare presentation of FCOD in an Indian middle aged male highlighting the diagnostic challenges.

Case report

A 38 year old male reported to the Oral medicine and Radiology department of with a chief complaint of swelling on both sides of lower back region of jaws since 8 months. History revealed the swelling gradually increased without pain. There were no significant medical and family histories. Patient was a chain smoker with 20 cigarettes per day for 20 years, but had discontinued it since seven months. Extraoral examination revealed facial asymmetry over posterior bilateral mandibular body and angle. The swellings were hard. Aspiration did not yield any significant material. On intraoral examination hard non tender both buccal and lingual cortical plates expansions in the molar regions. All associated teeth were vital. CBCT revealed mixed radiopaque and radiolucency with central radiopaque area encased by a diffused non corticated margin at the periapical area of 32 to 33 and 35 to 37 regions. There was evidence of bulbous roots with irregular deposition of cementum suggestive of hypercementosis with 35 and 36 (Fig1). Similar findings were noted on periapical region of 41 to 47 regions (Fig 2). A diagnosis of FCOD was made based on clinical and radiographic features with differential diagnosis of osteblastoma,

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ossifying fibroma and fibrous dysplasia.

Routine hematological values were within normal range and viral markers were negative. An incisional biopsy under local anesthesia was done and sent for histopathology.

Microscopy revealed a highly cellular fibrovascular connective tissue interspersed with numerous irregular curvilinear trabaculae of woven bone in 'ginger root' pattern and few globular calcifications resembling cementum (Fig 3, 4). Absence of osteoblastic rimming, typical Chinese letter pattern of bony trabaculae and presence of highly cellular stroma ruled out ossifying fibroma and fibrous dysplasia. Osteoblastoma was ruled out due to absence of vascular stroma and multinucleated giant cells. Lack of necrotic bone, inflammatory infiltrate negated chronic sclerosing osteomyelitis. A final histopathological diagnosis of FCOD was made.

As the patient was without any pain, surgical intervention was avoided. Patient was reassured about the clinical course of the condition and advised for follow up clinical and radiographic review every 6 months.

DISCUSSION

COD is a member of benign fibro-osseous disease. It was earlier classified as non-neoplastic idiopathic, reactive or hamartomatous lesion. Presence of cementum like histologic structure and association with apices of teeth, it was considered that it arises from apical periodontium. Lately, mutations of BRAF, HRAS, NRAS, FGFR3 and pathogenic hotspot mutations were noted in the RAS-MAPK pathway[4].

COD presents as 3 different forms depending on location and extent. PCOD at apical regions of mandibular anteriors, FCOD at periapex of single tooth in posterior jaws and FCOD as multiquadrant, multifocal involvement of jaw bones. FCOD more commonly occurs in middle aged African female (83.8%) and least in Asian female (1.6%). Gender wise, females more commonly affected (93.2%) than males (6.8%).[6] Mandible is more commonly involved than maxilla.[2,6] Our case occurring in a middle aged Asian male contributes the rarity.

Most of the reported cases were asymptomatic found incidentally on radiographs (85.3%).[2,3,6] our patient complained of bilateral swellings, an unusual finding as most of the symptomatic cases have been reported in older adults (>60 years) than asymptomatic ones in mean age of 51.2 years. [7-10]

Histopathologically, FCOD has to be differentiated from other osseous and cemental lesions, most commonly Ossifying Fibroma, Fibrous Dysplasia, Chronic Sclerosing Osteomyelitis , Complex Odontoma and Cementoblastoma.

Ossifying fibroma is a well-circumscribed, encapsulated neoplasm with higher cellularity and a storiform pattern. Often shows more prominent osteoblastic rimming.

Fibrous Dysplasia exhibits "Chinese character" woven bone trabaculae without osteoblastic rimming, blended into a more collagenous, less vascular stroma than COD.

Chronic Sclerosing Osteomyelitis characterized by dense, sclerotic bone with signs of chronic inflammation, such as inflammatory cell infiltration and irregular bone resorption, which is usually absent in primary COD.

Complex Odontoma shows a highly organized mixture of dental tissues (enamel, dentin, and pulp) that are typically absent in COD.

Cementoblastoma, a true neoplasm showing sheets of cementum-like material fused directly to the tooth root with a prominent rim of active cementoblasts.

As it is advised not to surgically intercept these lesions, lest complications such as poor healing, sequestrum formation etc, our patient is kept under follow up for observation.

CONCLUSION

Diagnosis of FCOD is to be done correlating clinical, radiological and histopathological observations and the other similar lesions to be differentiated as the treatment preferences differ in each lesion. Our case adds to the literature of FCOD in that it occurred in middle aged Asian male.

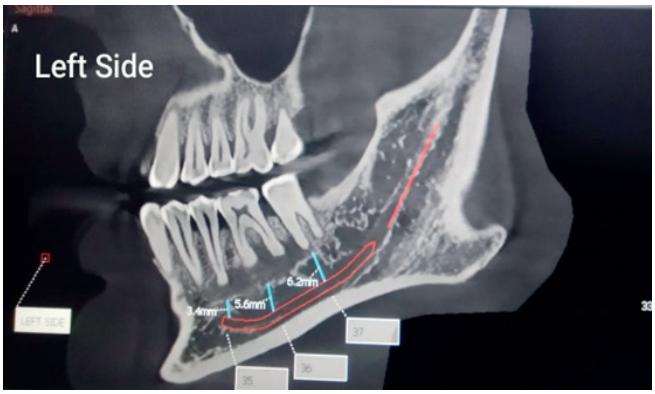

Fig 1: Mixed radiopaque and radiolucency at the periapical area of 35 to 37 regions.

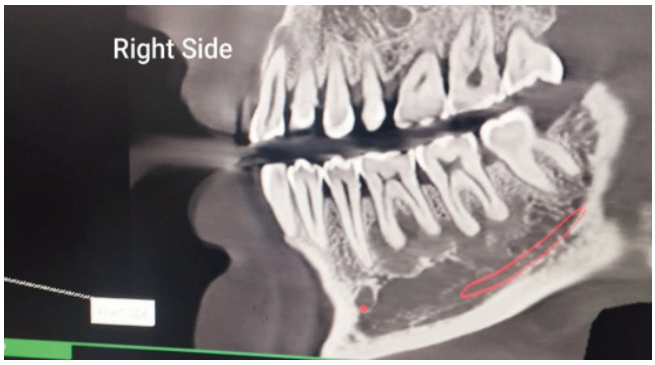

Fig 2: Mixed radiopaque and radiolucency at the periapical area of 41 to 47 regions.

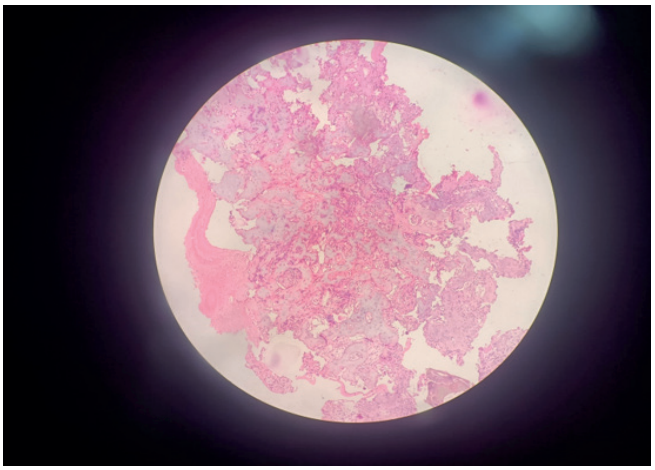

Fig3: Photomicrograph showing connetive tissue with numerous osseous trabaculae (H&E, 40x magnification)

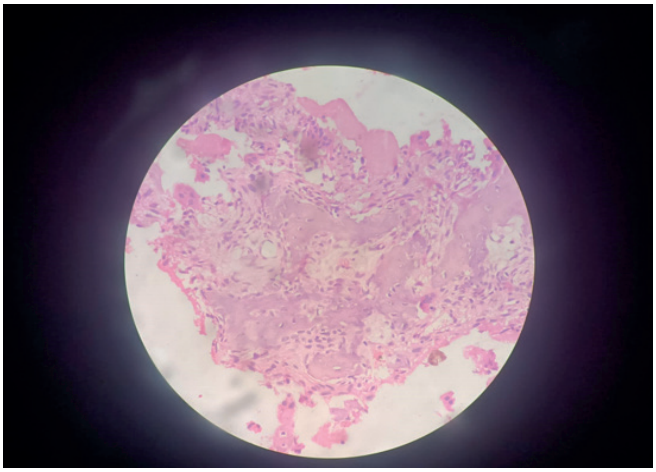

Fig4: Photomicrograph showing cellular fibrovascular connective tissue interspersed with numerous irregular curvilinear trabaculae of woven bone and few globular calcifications resembling cementum. (H&E, 100x magnification

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