FLORID CEMENTO-OSSEOUS DYSPLASIA – AN UNUSUAL ENTITY A CASE REPORT

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Abstract

Florid cemento-osseous dysplasia (FLCOD) is a rare, but well recognized condition that characteristically affects the jaws of middle aged women. It usually manifests radiographically as a diffuse, lobulated and irregularly shaped radio-opacities distributed throughout the alveolar processes which are usually bilaterally symmetrical. This condition has been classified as sclerosing osteitis, multiple enostoses, diffuse chronic osteomyelitis and gigantiform cementoma. The lesion is usually benign and requires no treatment unless cosmetically concerning or becomes symptomatic. For the asymptomatic patient the best management consists of regular recall examination with prophylaxis and maintenance of good oral hygiene According to the recent review of literature in 2006, only five patients of florid cemento-osseous dysplasia from India have been reported (less than 2%). Here we present such a rare case occurring in a 37 year old female who came with a swelling and dull pain in the lower right back teeth region since 1 and 1/2 years. Radiographically, it showed mixed radiolucent / radio-opaque mass in both right and left premolar to molar region.

Key Words: Fibro-Osseous Lesions; Florid Cemento Osseous Dysplasia.

Introduction

The term fibro–osseous lesion (FOL) is a generic designation of a group of jaw disorders characterized by replacement of bone by benign connective tissue matrix[1]. The classification of cemento-osseous lesions of the jaws has long been a matter of discussion for pathologists and clinicians. A review of the literature shows a wide range of terminologies used to describe what seem to be similar lesions[2]. In 1st Edition of World Health Organization (WHO) classification of Odontogenic tumors 1971, FLCOD was categorized under the Neoplasms & other tumors related to odontogenic apparatus[3]. The current classification of cementomatous lesions, released in 1992 by the WHO includes FLCOD under cemento – osseous dysplasia of the Neoplasms and other lesions related to bone [1,3]. This paper describes such a rare case diagnosed on the basis of clinical, radiographic and histological findings.

Case Report

A 37-year-old female patient presented to KM Shah Dental College & Hospital, Vadodara, with a complaint of swelling and dull pain in lower right back teeth region since 1 and ½ years. On extra-oral examination facial asymmetry was noted on lower right side of the face (Figure 1). Overlying skin was normal with no change in color, temperature and consistency. Intraoral examination revealed a localized swelling measuring about 2 X 1.5 cm extending from distal margin of right second premolar anteriorly to mesial margin of right third molar posteriorly. The mandibular vestibule was obliterated. On palpation, the swelling was hard in consistency. Teeth associated with the lesion were vital and the overlying gingiva and mucosa were normal.

Ortho pantomogram was taken and showed multiple sclerotic masses bilaterally in

tooth bearing areas of posterior mandible in relation to premolar and molar regions. Irregularly shaped radio opacities were distributed diffusely throughout the alveolar process enmeshed in poorly defined zones of decreased radio density. The borders of the lesion were indistinct and blended into the affected surrounding bone (Figure 2). Occlusal view showed no buccal or lingual cortical plate expansion (Figure 3). Posterior Anterior view of the skull showed mixed radio opaque and radio lucent areas on both sides of mandible. The left side of mandible was asymptomatic and apparently normal on clinical examination.

Surgical recontouring of the lesion on the right side was done for esthetic reasons. Multiple soft and hard tissue bits of varying sizes were received.

Microscopically, the H (hematoxylin) and E (eosin) stained sections showed fragments of soft and hard tissues. The soft tissue composed of cellular mesenchymal tissue with spindle shaped fibroblasts intermingled in thick bundles of collagen fibers and numerous small blood vessels. The hard tissue contained mixture of lamellar bone and globular shaped acellular, basophilic cementum like structures (Figure 4). Bony trabeculae were thick curvilinear resembling ginger root pattern (Figure 5). Few hemorrhagic areas were seen in close association with bony trabeculae. A diagnosis of florid cemento-osseous dysplasia was given.

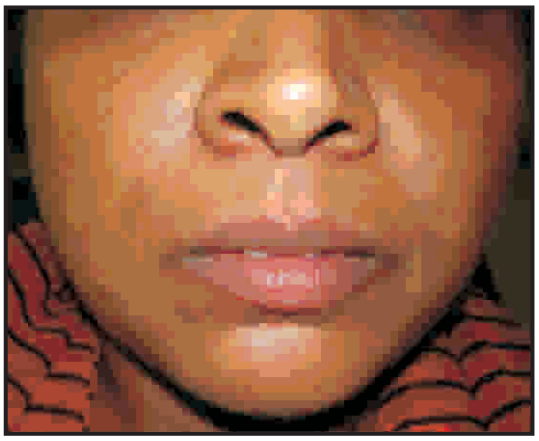

Fig 1: Clinical photograph

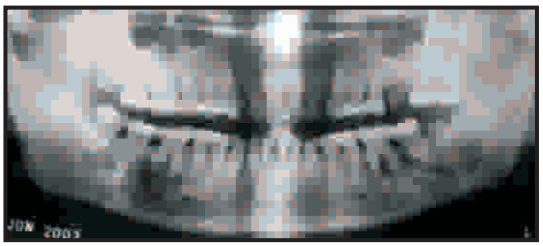

Fig 2: Ortho Panto-mograph showing multiple RO-RL masses in premolar and molar region of posterior mandible of left side

Fig4: Photomicrograph showing cementum masses and bony trabeculae in fibrous connective tissue [H &E]

Fig 5: Photomicrograph showing ginger root pattern of bony trabeculae [H& E, 40X]

Discussion

Cemento-osseous dysplasias are a group of disorders known to originate from periodontal ligament tissues and involve, essentially, the same pathological process. They are usually classified, depending on their extent and radiographic appearances, into three main groups: periapical (surrounds the periapical region of teeth and are bilateral), florid

(sclerotic symmetrical masses) and focal (single lesion) cemental dysplasias. Florid cemento-osseous dysplasia clearly appears to be a form of bone and cemental dysplasia that is limited to jaws. [4]

Florid cemento-osseous dysplasia was first described by Melrose et al. in 1976[2]. This condition has been interpreted as a dysplastic lesion or developmental anomaly arising in tooth-bearing areas.

FLCOD is more commonly seen in middle-aged black women, although it also may occur in Caucasians and Asians.[2,5] In some cases, a familial trend can be observed.[5] These lesions have a striking tendency toward bilateral symmetrical location, and it is not unusual to find extensive lesions in all four posterior (molar-premolar region) quadrants of the jaws. The radiographic appearance can vary from areas of radiolucency to mixed lesions and to opaque masses. Over the time the lesions tend to become increasingly radio opaque [6,7]. FLCOD blends with adjacent bone and therefore pathologists often receive multiple curetted fragments [8]. The lesions can be found in multiple quadrants in both the maxilla and mandible[6,9]. FLCOD is occasionally expansile and patients may experience pain rarely[9].

Histopathologically, FLCOD consists of dense, sclerotic masses that have been interpreted either as cementum or bone which are anastomosing and layers of cementum-like calcifications embedded in a fibro cellular background.[2,5]

Management of FLCOD may be difficult and not very satisfactory. When symptomatic, there may be pain alone or in association with sinus tracks with minimal purulent drainage.[10] Although in most situations the lesion is not treated, treatment is required when infection of the lesion occurs. For the asymptomatic patient the best management consists of regular recall examination with prophylaxis and maintenance of good oral hygiene.[11] Extensive surgical resection and saucerization are proposed treatment options when lesions become extensive and symptomatic.[12] Treatment of FLCOD with complete resection of the lesion would be impractical because it usually occupies most of the mandible and maxilla. When surgical intervention is indicated, a remodelling resection is recommended for esthetic reasons[5].

Chronic diffuse sclerosing osteomyelitis may resemble FLCOD, which appears as a single, poorly delineated opaque segment of the mandible. Clinically it involves the body of the mandible from the alveolus to the inferior border and may extend into the ramus. FLCOD 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 [5].

Only five Indian patients of florid cemento-osseous dysplasia have been reported (less than 2%), according to the recent review of recent literature.[12,13] This makes the occurrence of FCOD a relatively rare phenomenon. In the present case, no familial aspects of the disease were reported.

Florid cemento-osseous dysplasia is a self limiting disease restricted to alveolar processes. It is a distinct entity representing an exuberant variant of cemento-osseous dysplasia normally seen in multiple quadrants of both the jaws. Normally, a diagnosis of florid cemento-osseous dysplasia in the jaws is made by clinical findings, radiographic features and histology.[4] These lesions have to be included in the differential diagnosis of mixed radiolucent radio opaque lesions of the jaws.

Many lesions that occur in the jaw have a similar radiographical appearance and it is often difficult to differentiate among them. Differential diagnosis is crucial especially when there are coincidental findings like odontogenic infections or chronic diffuse osteomyelitis, neoplasia, bone dysplasia that can cause changes in the mandible with similar radiographic characteristics. Hence we must consider all the available diagnostic information, the radiographic characteristics which would contribute significantly to the diagnosis.

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