Multiple Odontogenic Keratocysts: A Case Report

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Abstract:

Multiple odontogenic keratocysts are a rare, but well recognized condition that characteristically affects the jaws of middle aged women. The radiographic appearance can vary from areas of radiolucency to mixed lesions and to opaque masses which are often bilaterally. 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, only five patients of florid cemento-osseous dysplasia from India have been reported (less than 2%). Here present such a rare case occurring in a 38 year –old- female who came with dull pain in upper right back teeth region since 6 months. 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 maxillofacial region is more prone to cystic lesions than any other part of the body and OKCs are the most common form of cystic lesions affecting the maxillofacial region (1). They are clinically aggressive lesions which are thought to arise from the dental lamina or its remnants (2). OKCs constitute about 3% - 21.5% of odontogenic cysts. (3-7). The peak incidence is during the second to fourth decades of life (7-10). Several studies indicate a male predilection (4-5, 10-12), some studies do not correlate with this (1, 8, 9). Majority of lesions occur in the mandible; mainly in the posterior body and ascending ramus. (5, 7-8). The angle (8) and symphyseal area is also frequently a locus for this lesion (13). The OKC can be very aggressive owing to its relatively high recurrence rate and its tendency to invade adjacent tissue (5). Therapeutic approaches vary from marsupialization and enucleation, combined with adjuvant cryotherapy or chemical cautery or Carnoy's solution to marginal or radical resection (9, 10, 14- 17). The recurrence rate has been reported to vary from 2.5% to 62.5% (5-8, 10, 14, 18,19). Malignant transformation of OKCs has also been reported (20). The high recurrence rate and aggressive behavior of the OKC have caused several investigators to regard it as a benign neoplasm rather than a cyst (7, 21). We discuss the possibility that the current case is a non-syndromic presentation of multiple OKCs.

CASE REPORT

A 38 year-old- female patient reported to the Dental Clinic, complaining dull, intermittent and non-radiating type of pain in right upper and lower back tooth region since 6 months. Pain was aggravated on lying down and was relieved by taking medications. On intra oral examination there was no evidence of dental decay or periodontal disease. While tender on

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percussion was elicited i.r.t 14,15,16,17,44,45,46,47 .Pulp vitality showed that all the concerned teeth was vital, so with all these clinical findings initially chronic apical periodontitis was suspected.

Then patient was subjected to Panoramic radiography which revealed multiple irregular radiolucencies, interspread by multiple radiopacities in tooth bearing areas of posterior mandible in relation to premolar and molar regions bilaterally . The borders blended into the affected surrounding bone. The lesion was fused to involved teeth but there was no root resorption. Similar lesions were also found in both quadrants of maxilla, but appeared to be less homogenous. (fig.1)

Fig.1- Panoramic radiograph

Further to precisely determine the extent of lesion, Non Contrast Computer Tomography (NCCT) was done which revealed large radiolucent shadows extending in both maxilla and mandible. The lesion occupied all the four quadrants of the jaw and consisted of radiolucent masses with unclear radiopaque margin (Fig 2).

Fig 2: Non Contrast CT Scan

With the help of clinical and radiographic investigations the diagnosis was made as Multiple Odontogenic Keratocysts. Unicystic ameloblastoma, adenomatoid odontogenic tumor and central giant cell granuloma were considered under differential

diagnosis. Complete blood count and other laboratory data were within normal limits.

The patient was then referred to the department of General Medicine for evaluation. Nothing abnormal was detected on review of systems. However dermatologic consultation revealed acanthosis nigrans. The patient was then referred to the department of Oral Surgery. Under general anesthesia, the lesions were enucleated under general anesthesia. During the surgery, white cheesy material was found extruding from the cystic lesion.

DISCUSSION

Cemento-osseous dysplasias are a group of disorders known to originate from periodontal ligament tissues and involve, essentially, the same pathological process.⁵ 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 dysplasias¹. Florid cemento- osseous dysplasia clearly appears to be a form of bone and cemental dysplasia that is limited to jaws⁵.

Florid cemento-osseous dysplasia was first described by Melrose et al. in 1976.⁶ This condition has been interpreted as a dysplastic lesion or developmental anomaly arising in tooth-bearing areas.

Melrose et al. reported a study of 34 cases of such lesions, of which 32 were black women (in a predominantly Caucasian population) with a mean age of 42⁶ and Mac-Donald also reported sixteen cases of FLCOD (mean age, 52.1 years) of which six were West Indian women and ten were Chinese.⁷

Only five Indian patients of florid cemento-osseous dysplasia have been reported (less than 2%), according to the review of recent literature. This makes the occurrence of FCOD a relatively rare phenomenon^{8,9,10}.

FLCOD is more commonly seen in middle-aged black women, although it also may occur in Caucasians and Asians.² In some cases, familial trend can be observed¹¹.

In the present case, no familial aspects of the disease were reported .These lesions have a striking tendency toward bilateral symmetrical location and it is usual to find extensive lesions in all four posterior (molar-premolar region) quadrants of the jaws.^{4,12} Symptoms such as dull pain or drainage are almost always associated with exposure of sclerotic calcified masses in the oral cavity .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 radiopaque.^{12,13} in our case the

lesion is mixed means it is in the early stage. FLCOD blends with adjacent bone and therefore pathologists often receive multiple curetted fragments.¹⁴ FLCOD is occasionally expansile (not expansile in our case) and patients may experience pain rarely.¹⁵

Radiologically, it must be differentiated from other lesions such as Paget's disease, sclerosing osteomyelitis, periapical cemento osseous dysplasia (PCOD), focal cemento-osseous dysplasia (FCOD) and multiple idiopathic osteosclerosis.

PCOD and FCOD have several similarities with FLCOD including age, sex and racial profiles of the patients and comparable radiographic and histologic appearances. However, FCOD is a single lesion compared with FLCOD that consists of multiple lesions. PCOD usually involves the anterior segment of the mandible, but FLCOD usually involves three or four quadrants.¹⁶

Idiopathic osteosclerosis is a well-defined lesion without a radiolucent rim that has a bony internal structure, whereas FLCOD has a cemental internal pattern. Idiopathic osteosclerosis also occurs as multiple lesions in patients with colorectal carcinoma or adenoma and Gardner syndrome.¹⁷

Paget's disease of the bone may have a cotton-wool appearance. However, this condition affects the bone of the entire mandible, while florid cemento-osseous dysplasia is placed above the inferior alveolar canal. Paget's disease shows loss of lamina dura, but lamina dura is not affected by FLCOD. Paget's disease is often polyostotic, involving other bones such as the spine, femur, skull, pelvis and sternum and leads to biochemical serum changes, such as elevated alkaline phosphatase levels. No involvements of other bones were found in these reported cases.¹⁸

Chronic diffuse sclerosing osteomyelitis is another disease that should be differentiated from FLCOD. It appears as a single, poorly delineated opaque segment of the mandible, whereas florid cemento-osseous dysplasia is seen as multiple round or lobulated opaque masses. Chronic diffuse sclerosing osteomyelitis involves the body of the mandible. In addition, FLCOD is frequently associated to black women, while chronic diffuse sclerosing osteomyelitis does not generally occur in women.^{2,19}

Management

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.²¹ 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.²² Extensive surgical resection and available diagnostic information, the saucerization are proposed treatment options when lesions become extensive and symptomatic. 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.¹¹

Conclusion

In conclusion, generally when a lesion is found in the jaws, the diagnosis is suggested by clinical and radiographic findings and is confirmed by histopathology. However, FLCOD is a condition in which the diagnosis relies on radiology and clinical findings alone, and biopsy is not mandatory due to the increased risk of infection.

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