Florid Cement Osseous Dysplasia and Chronic Diffuse Osteomyelitis in Maxilla

Mahnaz Saheb-Jamee,1 Fateme Arbabi-Kalati1,2

1. Department of Oral Medicine, Faculty of Dentistry, Tehran University of Medical Sciences, Tehran, Iran
2. Department of Oral Medicine, Genetics of Non Communicable Disease Research Center, Faculty of Dentistry, Zahedan University of Medical Sciences, Zahedan, Iran

Abstract

Chronic diffuse osteomyelitis is an intermodular bone infection which may be resulted from a localized osteitis, previous acute osteomyelitis or prior infective processes. Florid cement-osseous dysplasia brings about a change in perfusion and in case of infection will pave the way for osteomyelitis. Complaining of chronic pain and pus drainage, a patient referred to the center of oral diseases for removing the right maxillary first molar and was diagnosed with chronic diffuse osteomyelitis in florid cement-osseous dysplasia. Bone expansion of all four quadrants of the jaws, pain, pus drainage, sinus involvement made this patient unusual (abnormal).

Introduction

Benign fibro-osseous lesions refer to a group of disorders in which the bone is replaced with fibrous connective tissue containing abnormal bone or cementum. According to the WHO definition, these lesions are divided into 3 groups including florid cement-osseous dysplasia (FLCOD), periapical cemento-osseous dysplasia and focal cemento-osseous dysplasia [1].

FLCOD was first defined by Melrose et al. in 1976. In usual radiographies this lesion can be observed as multiple sclerotic lesions with upper lucent margin of the inferior alveolar canal. Histologically, lesions contain immature osseous trabeculae and cementum-like calcifications which are buried in fibroblast. Middle-aged black women are affected (involved) more than others, although the lesion involves Caucasians and Asians, too. Although family involvement is reported in a few cases, but these lesions don’t have hereditary basis. Certain biochemical changes are not reported with the disease [2-4]. The process of this lesion may be completely asymptomatic which in these cases they will be detected in radiographies that are performed for other reasons. The lesions are usually bilateral and symmetrical and the involvement of all four quadrants of the jaws is not unusual. Chronic diffuse osteomyelitis is an intermodular bone infection which may be resulted from a local osteolysis, pre-existing acute osteomyelitis or pre-existing infective processes. Jacobson has described clinical, diagnostic perspectives and treatment of chronic diffuse osteomyelitis.

Case Report

In 2009, a 42-year-old woman complaining of pain and pus drainage in maxillary right molar referred to the department of oral diseases, Faculty of Dentistry, Tehran University of Medical Sciences. In the patient’s history, no systemic disease or family record of similar illnesses was mentioned. In intraoral examination, bone expansion was observed in buccal and lingual area of maxillary right molar (Fig. 1). Also, there was slight bone expansion in other jaw quadrants. According to the patient, bone expansion was started 2 years ago and pain and pus drainage created four months ago after extracting (pulling) the infected tooth from maxillary right molar (Fig. 2).

Periapical and panoramic radiographies indicated several sclerotic masses with frosted glass (opaque) perspective and with a rim lucent in mandible and maxilla. In the CT-scan, the involvement of right maxillary sinus was specified (Fig. 3).

Histological examination indicated segments of necrosed bone and cementum-like materials. Bacterial colonies, neutrophil accumulation and granulation tissue was observed around the bone segments. Since Paget’s disease was discussed in differential diagnosis, biochemical evaluation of alkaline phosphatase, calcium and phosphorus was done and its quantities (amounts) were at normal range.
Discussion

The term “florid cemento-osseous dysplasia” was replaced with gigantiform cementoma in WHO’s second review of odontogenic tumors international classification. The disease is restricted to areas with jaw teeth (alveolar region) and is not associated with another bone disease. In usual radiographies, these lesions have sclerotic perspective and the existence of this disorder will lead to decrease in bone perfusion and will predispose patients to osteomyelitis. In differential diagnosis, the lesion was recognized as Paget’s disease but the disease involves the whole jaw and will result in loss of lamina dura. Involvement of several bones and increase of alkaline phosphatase is its characteristics but FLCOD is upper the alveolar inferior canal and lamina dura will remain intact. Involvement of other bones and biochemical changes doesn’t exist. Based on clinical manifestations and biochemical tests Paget’s disease was rejected [5]. FLCOD may be inherited as autosomal recessive but few cases of familial incidence of the disease is reported; our patient didn’t mention family history of similar lesion, too. FLCOD is a benign disease and the patient must only be under radiography and clinical follow-ups but when there is infection, it will result in osteomyelitis due to poor perfusion of bone and then invasive antibiotic therapy and surgery must be regarded in treatment plan [5].

The patient was referred to oral and maxillofacial surgeon for continuation of treatment. Due to the spread of infection and sinus involvement, the patient was treated with maxillectomy. In a report, for treatment of a similar case of infectious and invasive FLCOD, mandibulectomy and common alveolar reconstruction has been done in a 47-year-old black woman [1].

The important point about this patient is his dentist’s negligence and not preparing radiography before pulling the tooth which leads to improper treatment, causing an additional trauma, infection and finally defectiveness of the patient

Authors’ Contributions

All authors had equal role in design, work, statistical analysis and manuscript writing.
Conflict of Interest
The authors declare no conflict of interest.

References

Funding/Support
Tehran University of Medical Sciences