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Osteomyelitis mimicking Paget's disease or a pagetoid phenomenon : a case report

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Summary
Osteomyelitis is an inflammatory infection of the bone marrow. When the maxillofacial region is involved the mandible is the preferred site in adults and the elderly. Maxillary involvement is usually seen in children with only few cases reported in adults. The disease occurs in stages, with some stages highly mimicking other diseases. Clinicians ought to be aware of the full biological course of osteomyelitis of the jaws to avoid mis-diagnosis or underdiagnosis.

Keywords: *Maxillary osteomyelitis, paget's disease of bone, pagetoid phenomenon and alkaline phosphatase.*

Résumé
L'osteomyelite est une inflammation de la moelle osseuse. Lousque la region maxillofaciae est implique, la mandibule est le. Site de predilection chez les adultes et les personnes ages. Le maxillaire est souvant implique chez les enfants. Quelques cas ont été repporte chez les adultes. La maladie invovient par etapes. Certains etapes invovient d'autres maladies. Les clinicians doivent connaitre toute l'evolution biologique de l'osteomyelite des joues pour eviter des eveuns diagnostiques.

Introduction

The skeletal tissues, like the soft tissues of the body, respond to injuries by a process of inflammation. In many cases the injurious agent is of bacterial nature, as in osteomyelitis caused by staphylococci in most cases, and various streptococci or mixed organisms in other instances. Studies suggest osteomyelitis to be predominantly a disease of the adult, and the elderly, particularly the chronic suppurative and sclerosing types [1,2].

Unlike osteomyelitis, Paget's disease of bone (osteitis deformans) is a common chronic osteolytic and osteosclerotic disease of uncertain cause. There is a view [2] that a viral agent might be involved. The disease is however similar to osteomyelitis in the afflicted age group, which is 40 years and above.

The diagnoses of Paget's disease of the bone and osteomyelitis could sometimes be difficult, as each of the entities develop in stages: with some stages clinically and histologically mimicking each other. Both diseases have also been found co-existing with and complicating other diseases [3,4]. Very few reports exist in the literature on osteomyelitis of the jaws among Nigerians. The thrust of this report is to highlight the possible diagnostic difficulties, which chronic sclerosing osteomyelitis can pose, and to be aware of its full clinical and biological course, especially in the maxillofacial region.

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Case Report

A 70 year old elderly woman presented on the 30th of December 1997 to the oral diagnosis clinic of the University College Hospital Dental centre. She had a history of dull pain, swelling and suppurative discharge around the gingival crevices of left upper 2nd premolar and 2nd and 3rd molar teeth. Her past medical history was not significant, and her dental history revealed previous extractions of some upper right molar teeth which was uneventful.

Extra-oral examination showed a slight swelling of the left maxillary region with left sub-mandibular lymphadenitis. Intra-orally, she had a mobile left upper 2nd premolar and 2nd and 3rd molars with buccal swelling, and discharge around the surgical crevices of the mobile teeth. Initial clinical impressions made included; Paget's disease of the maxilla, fibrous dysplasia and chronic osteomyelitis of the maxilla.

Patient was referred for occipitomental and oblique occlusal radiographs. The occipitomental view showed a dense opacity of the left antrum, while the oblique occlusal radiograph of the maxilla showed a cotton wool appearance. Further investigation included blood chemistry which showed Ca^{++} - 9.6 mmol/L, and PO_4 – 5.6 mmol/L. Alkaline phosphatase was 168 iu/L. Culture of pus exudate from the crevices of the attending teeth was sensitive to tetracycline.

Patient was covered with tetracycline 500mg 6 hourly for two weeks, during which time the mobile left upper 2nd premolar and 2nd and 3rd molar teeth indicated were extracted, along with an incisional biopsy of the supporting bone. Histopathology report indicated a diagnosis of chronic diffuse sclerosing osteomyelitis. Patient was subsequently seen on follow-up, and there was evidence of progressive resolution of the swelling for about two months after which the patient defaulted.

Discussion

Osteomyelitis is not an uncommon disease. Therefore this case has been reported not because of its rarity, but due to its the unusual mode of presentation with radiographic features suggestive of Paget's disease of the bone. The disease can be encountered at any of its three stages: acute, sub-acute, and chronic suppurative or chronic sclerosing. Diagnosis is frequently made on the basis of the clinical findings from the initial stages of the disease that are correlated with radiographic, laboratory, and histopathologic data collected as the disease progresses. In the chronic sclerosing or suppurative stages, it has typical radiographic, and histological features that mimic other facial swellings particularly Paget's disease of bone.

Paget's disease, of bone like chronic sclerosing osteomyelitis is usually seen in patients above age 40 years, with a male predilection. The jaws are frequently affected, with the maxilla more frequently being affected than the mandible. Maxillary osteomyelitis is quite rare in adults [1,2]. Koorbusch *et al.* [1] reported only two cases of maxillary osteomyelitis out of 35 cases reviewed over many years, while Daramola and Ajagbe [5] reported none in the elderly group in their 34 cases. Hence the choice of Paget's disease of the bone as a possible differential diagnosis in this case was a sound clinical decision, even though Paget's disease of the bone is considered to be very rare in the black population [2]. Futhermore, the suppuration encountered in our patient even though not a typical feature in Paget's, disease of bone can be brought about by a super-imposition of chronic periodontal disease, which the patient was noticed to have had. Other features noted were jaw enlargement with teeth mobility. These also are common to both osteomyelitis and Paget's disease of bone.

The radiographic findings in Paget's disease of the bone and osteomyelitis depend on the stage of the disease. In the early stages of Paget's disease, irregular radiolucency is evident but progressively turns to radio-opacity and finally, dense irregular radiopaque areas resembling "cotton wool".

Fig 1: Occlusal radiograph of patient showing a typical 'cotton wool' appearance

The occlusal radiograph of the patient (Fig. 1) in our opinion met the radiographic criteria for a late stage Paget's disease of the bone. Many authors [2,6] consider the 'cotton wool' appearance as almost pathognomonic for Paget's disease of the bone; they however, advised caution on rushing to a diagnosis as osteomyelitis and Paget's disease might be indistinguishable radiographically at certain stages.

Histological examination is usually the final arbiter in clinical diagnosis. Bone in chronic osteomyelitis may be thickened and contain prominent 'cement lines' which may mimic Paget's [2] disease (Fig. 2). In many studies [2] the cement line is accepted as the main histological feature that separates Paget's disease of the bone from other osteolytic and osteoblastic lesions.

Fig 2: Histologic section of bone specimen from patient's maxilla showing 'cement lines' x 100

For any clinician therefore, who might not have a full complement of radiographic, histologic, and other pertinent laboratory investigations at his/her disposal, diagnosing the late stage of chronic sclerosing osteomyelitis of the maxilla might prove difficult. Cabanela *et al.* [7], and Jacobson *et al* .[8] in their studies have highlighted the potential for osteomyelitis to mimic other lesions, thus leading to a mis-diagnosis.

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