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CHRONIC FOCAL SCLEROSING OSTEOMYELITIS OF MANDIBLE – A CASE REPORT

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ABSTRACT

The term osteomyelitis encompasses a broad group of infectious diseases characterized by infection of the bone and/or bone marrow. The pathogenesis of these diseases can follow acute, subacute or chronic courses and involves a range of contributory host and pathogen factors. A commonly used aetiological classification distinguishes between three types of osteomyelitis: acute or chronic haematogenous disease seeded by organisms in the bloodstream, local spread from a contiguous source of infection and secondary osteomyelitis related to vascular insufficiency. Risk factors, such as systemic or local immunocompromise and prosthetic implantation, can increase one's risk for infection. The unique demarcated environment of osteomyelitis results in a high-grade local inflammatory host response leading to formation of sequestrum. This report discus a case of chronic focal sclerosing osteomyelitis with a history of drug therapy leading to an immunocompromised state systemically.

Keywords: Osteomyelitis , Bone Scleroses, Sequestrum.

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INTRODUCTION

The term sclerosing osteomyelitis is applied specifically to those lesions which have formed directly as a result of infection. This term is synonymous with chronic productive osteitis, chronic focal sclerosing osteomyelitis, and chronic local sclerosing osteomyelitis. This type of sclerosis is confined to the limits of the bone and does not result in any expansion of the surrounding cortex. Bone sclerosis is used as a general term which refers to increased bone formation resulting in an increased radiopacity in an area [1]. An osteosclerotic area is usually localized, abnormally opaque, often round or elliptical, and may vary from a few millimeters to several centimeters in size. The outline is usually reasonably distinct, and overall the area may be either uniformly radiopaque or patterned to give a 'ground glass' or 'stippled' appearance.

An uncommon, but often distressing clinical situations is encountered in patients with large, dense, sclerotic masses within the jaws. Pain is a common complaint, and fistulas with minimal purulent drainage may be presenting symptoms. In other instances, however, radiologically identical lesions are asymptomatic and are discovered during the course of a routine radiographic examination. Lesions of this type have been designated as focal or diffuse chronic sclerosing osteomyelitis [2], sclerosing osteitis [3], multiple enostosis [4], sclerosing cementoma [5] and gigantiform cementoma[6].

Radiographically, the lesions present as dense, structure less, radiopaque masses with fairly well delineated margins, symptoms are variable , but when the lesions becomes exposed to the mouth, pain and purulent drainage may result.

This case report discusses the case of focal sclerosing osteomyelitis of mandible with a history of a drug induced immunocompromised state of the patient.

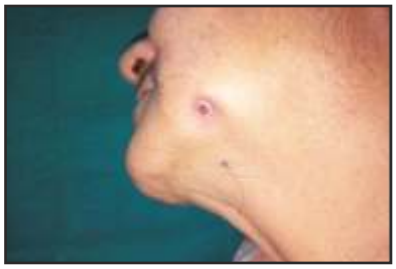

Figure 1 : Extra-oral Patent Sinus Track

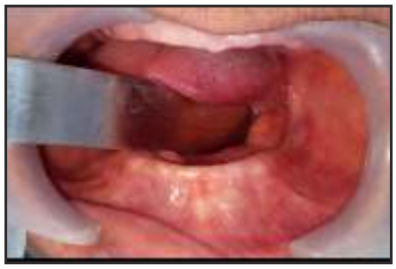

Figure 2 : Complete Edentulous Mandibular Ridge

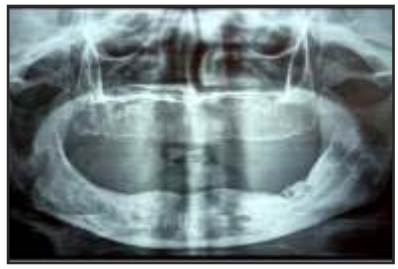

Figure 3: Ortho Pantomograph – Revealing A Radio-opacity In 37 Region

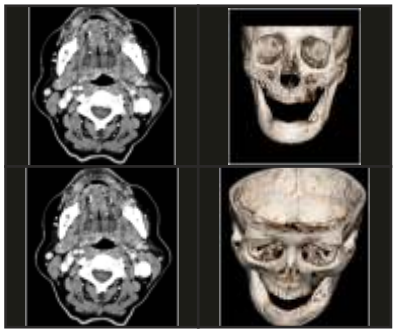

Figure 4: CECT Mandible

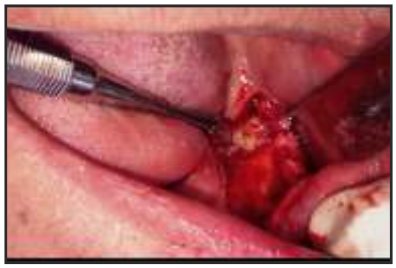

Figure 5: Bony Sequestrum In 37 Region

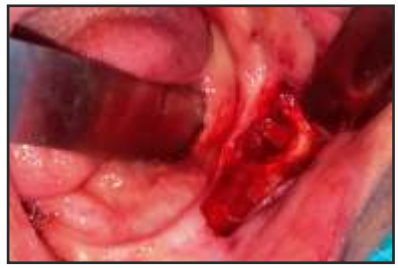

Figure 6: Sequestromy Followed By Curettage

Figure 7: Soft Tissue And Hard Tissue Sent For Histopathological Examination

Figure 8: Histopathological Slides- Soft Tissue & Hard Tissue (10x)

CASE REPORT

A 60-year-old female, presented in the department of oral and maxillofacial surgery, Sri Guru Ram Das Institute of Dental Sciences and research, Amritsar, with a complaint of pain in left posterior mandibular region, along with a discharging sinus at lower border of mandible(Fig :1). The skin around the sinus region was fibrous with firm adherence to underlying tissues. On intra-orally examination, patient had a complete edentulous mandibular ridge with normal overlying tissue (Fig: 2).

An Ortho-pantomo graph revealed a radio-opacity of the size approximately 1 cm x 1cm in the 37 tooth region, suggestive of either a sequestrum or a sclerotic bony fragment (Fig: 3). Contrast Enhanced Computed Tomography (CECT) reported generalized increased density in the marrow of the mandible in the region of the angle, adjacent body and ramus on the left side with some degree of cortical thickening with irregular increased density/ sclerotic focus surrounding by lucency along superior alveolar margin of mandible in the region suggestive of sequestrum in nature, with ill defined hypodensity in the adjacent soft tissue with a track extending along subcutaneous fat upto skin surface suggesting a sinus track(Fig :4).

Blood investigations were advised in which total leucocyte count (TLC) was 8200/mm^3, erythrocyte sedimentation rate (ESR) came to be 42mm/hour and C-reactive protein (CRP) was 29mg/L. All these blood finding are indicative of chronic osteomyelitis.

Patient gave a history of arthritis since 5-6 years for which she was taking glucosamine (500mg-tds). After thorough evaluation of clinical, radiological and hematological findings, a provisional diagnosis of osteomyelitis was made and a surgical intervention was planned for the patient.

Surgical procedure

Locally anesthesia 2% lignocaine with 1:200,000 epinephrine was administered. Crestal incision was given in left mandibular edentulous ridge staring from 33 region till 38 region. Mucoperiosteal flap was raised (Fig: 5). Sequestromy followed by curettage of the surgical bed was done, during which a bony sequestrum of around 1.5 cm and 1.5 cm as removed from the region of 37(Fig :6). Granulation tissue in the region of 37 was excised using a curette. After compete curettage, copious irrigation with normal saline and povidine

iodine was done. Primary closure was done using 3-0 silk sutures. Patient was started on regular antibiotic regime (amoxicillin plus clavulanic acid-625 mg and metranidazole-400mg). The soft tissue and hard tissue were sent for histopathological examination (Fig: 7).

Patient was recalled on 7th post operative day for follow up. The skin and subcutaneous tissue over the sinus area had lost its adherence to underlying bone. Extraoral sinus was healed.

Histolpathological evaluation revealed a fragment of devitalized bone which lack osteoblastic rimming. Osteocytic lacunae are empty. Soft tissue bit shows a moderately inflamed connective tissue stroma with hemorrhagic areas. All these features were suggestive of chronic focal sclerosing osteomyelitis (Fig: 8).

DISCUSSION

Osteomyelitis is a progressive infection of bone [7] that results in inflammatory destruction of the bone, bone necrosis, and new bone formation and may progress to a chronic and persistent state. The major categories of osteomyelitis are based on the source of infection (hematogenous or secondary to a contiguous focus of infection) and whether vascular insufficiency (either local or systemic) exists[8]. While large-organism inoculation and/or host compromise can predispose patients to the development of osteomyelitis, the virulence of the infecting pathogen also has a significant role. One species in particular, *Staphylococcus aureus*, is able to cause an acute bone infection even with a low inoculum in a healthy host. In addition, through the timed expression of its arsenal of virulence factors and aided by its ability to develop antibiotic resistance rapidly, *S. aureus* progresses to a chronic, biofilm-mediated infection. Once a chronic infection develops, bacterial clearance cannot be attained by the host immune system or antimicrobial therapy. At this point, surgical removal of the nidus of infection is usually necessary for complete infection resolution.

Predisposing factors to osteomyelitis can be fractures due to trauma and road traffic accidents, radiation damage, paget`s disease, osteoporosis, systemic diseases like malnutrition, acute leukemia, uncontrolled diabetes, sickle cell anemia, chronic alcoholism or any drug therapy. Etiology behind all these pathologies is disruption of vascular supply in the nutrient arteries.

In the literature, many authors suggest an infectious origin of focal sclerosing osteomyelitis. Microorganism usually involved is Staphylococcus aureus and *S. epidermidis*

In this report, the case discussed has all the contemplating features of chronic focal sclerosing osteomyelitis. Clinical finding like presence of the patent sinus track since 4-5 months associated with mild to moderate pain in the area, including the radiological outcomes such as increased density in the trabeculae patter of the mandible in the region of the angle, with increased degree of cortical thickening. Sequestrum as a sclerotic focus is seen around the superior alveolar margin of mandible.

The important aspect of the differential diagnosis of the case is history of use of glucosamine, an amino sugar, by the patient since 5-6 years. A specific polysaccharide antigen named polysaccharide intercellular antigen (PIA) was isolated. PIA is composed of *N*-acetyl glucosamine residues (80 to 85%) and an anionic fraction with a lower content of non-N-acetylated D-glucosaminyl residues that contains phosphate and ester-linked succinate (15 to 20%) [9]. PIA is a polymer of approximately 130 residues, but other sizes of this N-acetylglucosamine have been identified, termed PNAG-I (the immunogenic 460-kDa compound), II (100 kDa), and -III (21 kDa) [10]. PIA is produced in vitro from UDP-acetylglucosamine via products of the intercellular adhesion (*ica*) locus [11]. The genes and products of the *ica* locus have been demonstrated to be necessary for biofilm formation and are upregulated in response to anaerobic growth, such as the conditions seen in the biofilm environment [12]. Biofilm formation by this pathogen and other microbial species are responsible for osteomyelitis allowing immune evasion, as well as resistance to clearance by antimicrobial agents. After the accumulation of microorganisms, infection in the bone leads to an increase in intramedullary pressure due to inflammatory exudates. The periosteum becomes stripped from the osteum, leading to vascular thrombosis. Bone necrosis follows due to lack of blood supply and lead to the formation of sequestrum.

Osteomyelitis is an infectious process with a spectrum of clinical demonstrations from a localized small sclerosing lesion to a generalized suppurative destructive one. It represents the interactions between host immunity and microbial virulence [13]. With a fully competent immune defense, intact osseous tissue is able to restrict the infectious pathogens or kill them [14]. Remodeling capacity of the involved bone has an important role in reactive bone formation following the inflammatory processes [15]. These reactive calcifications may show snowflake, cotton-wool or even sun ray appearances. It is a critical rule that traumatic, infectious and benign reactive lesions are more common than malignant pathoses in the oral cavity. Osteomyelitis may share some radiographic features with primary or metastatic malignancies of the jaw[16].

CONCLUSION

This case report demonstrates the typical features of chronic focal sclerosing osteomyelitis, a rare but well-described potential complication of chronic odontogenic infections that dentists may more frequently encounter. Management entailed a course of antibiotics in combination with surgical debridement. Some reports have also advocated the use of hyperbaric oxygen in the treatment of this condition, especially in the irradiated mandible. In the present case, the patient was prescribed a two week course of oral amoxicillin plus clavulanic acid-625 mg and metranidazole-400mg, which, in combination with surgical debridement was successful.

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