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CASE REPORT

Actinomycotic Osteomyelitis of Mandible Masquerading Periapical Pathology

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ABSTRACT

Actinomycosis is an infectious disease characterized by chronic granulomatous and suppurative lesions often caused by Actinomyces group. Actinomycosis has been referred to as the chameleon of head and neck pathology because of its varied clinical picture which can resemble various pathologies ranging from benign infection to metastatic tumour. Nowadays, the diagnosis of the same may be difficult because the number of patients with typical symptoms has decreased, and there is a low success rate in culturing the microorganism. Mandibular osteomyelitis is also underappreciated by many clinicians in their assessment of head and neck infections. Most cases are traced to an odontogenic source, with periapical tooth abscess and posttraumatic or surgical complication as key antecedent events.
A case of osteomyelitis of mandible initiated from a vital but peridontally compromised tooth is reported. Radiography of this case revealed bone destruction, and H & E of the biopsied tissue showed branching filaments resembling ray fungus.

Keywords: Actinomyces, Osteomyelitis, Filamentous, Ray fungus

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INTRODUCTION

Von Langenbeck noted the first case of human actinomycosis in 1845, and attributed it to a fungus. Bollinger described the organism *Actinomyces bovis* and its ability to cause "lumpy jaw" in cattle. The word Actinomyces means "ray fungus," and reflects the general belief at the time that the organism was a fungus. The organism was first isolated from humans in 1891, when Wolff and Israel reported culturing it anaerobically and growing only at body temperature. Actinomycosis is a Greek word comprising of "Aktino" meaning radiating appearance of sulfur granules and "mykos" which labels the condition as mycotic disease. In the 1960s, Waksman concluded that Actinomyces was actually a gram-positive bacteria (1).

Actinomycosis is a sub acute to chronic bacterial infection caused by filamentous, gram-positive, non-acid fast, anaerobic to microaerophilic bacteria. It is characterized by contiguous spread, suppurative and granulomatous inflammation, and formation of multiple abscesses and sinus tracts that may discharge sulfur granules. The most common clinical forms of actinomycosis are cervicofacial (i.e., lumpy jaw), thoracic, and abdominal. In women, pelvic actinomycosis is also possible (2).

Actinomycosis is an infectious disease that is characterized by chronic granulomatous and suppurative lesions often caused by Actinomyces israelii. Actinomyces infection had occurred in the cervical region in approximately one half of the patients in several series as reported by Smith and Mc Quarrie et al, with soft tissue being more commonly involved than bone(3). Incidence of Actinomyces infection affecting Mandible is (53.6%), Cheek (16.4%), Chin (13.3%), Maxilla (5.7%) and TMJ (0.3%). Osseous involvement occurs only in 15% of cases.

PATHOPHYSIOLOGY

Actinomycetes are prominent among the normal flora of the oral cavity but less prominent in the lower gastrointestinal tract and female genital tract. Because these microorganisms are not virulent, they require a break in the integrity of the mucous membranes and the presence of devitalized tissue to invade deeper body structures and to cause human illness (4). Furthermore, actinomycosis is generally a

polymicrobial infection, with isolates numbering as many as 5-10 bacterial species. Establishment of human infection may require the presence of such companion bacteria, which participate in the production of infection by elaborating a toxin or enzyme or by inhibiting host defences. These companion bacteria appear to act as co pathogens that enhance the relatively low invasiveness of actinomycetes. Specifically, they may be responsible for the early manifestations of actinomycosis and for treatment failures (4).

It has been proved that Actinomyces requires the presence of many other types of bacteria to proliferate; the specific ecosystem thus formed has a low oxido-reduction potential that is favourable to anaerobic growth. This ecosystem is formed of a poly microbic "associate" flora working in synergistic fashion. It destroys local tissue in a highly vascularised, and therefore aerobic region and replaces it with a poorly irrigated granulation tissue, thereby permitting the development of an anaerobic milieu that is the key to Actinomyces development (5).Once infection is established, the host mounts an intense inflammatory response (i.e. suppurative, granulomatous), and fibrosis may then follow. Infection typically spreads contiguously, frequently ignoring tissue planes and invading surrounding tissues or organs. Ultimately, the infection produces draining sinus tracts. Haematogenous dissemination to distant organs may occur in any stage of actinomycosis, whereas lymphatic dissemination is unusual (4).

CASE REPORT

The case was a 43 year old man who had low intensity pain in left lower jaw since one month. He consulted his general dental practitioner for pain and took an antibiotic medicine for which he had no record. Pain was relieved but a solitary sinus tract opening in the region of 35 and 36 appeared introrally after the antibiotic therapy. He, as advised by his general dental practitioner underwent the extraction of 35 and took multiple antibiotic therapies, but was left without cure. The fluid coming out of the sinus was frank pus and there was no relief in the sinus drainage (Fig. 1). The solitary sinus persisted after extraction and the patient had developed the fear of growing carcinoma in that area since the problem was not solved. With this apprehension he reported to the Government Dental College, Rohtak. On intra oral examination 36 was vital with deep pocket and furcation involvement (Fig.2). The Intra oral peri-apical radiograph with Gutta-percha inserted into the sinus opening revealed a big radiolucency in relation to 36 & 37 and to outline the lower border of radiolucency a Panoramic radiograph was also taken (Fig.3).

Since there was a deep pocket and furcation involvement in relation to 36 & 37, a flap surgery was planned. On raising the flap, the furcation area was curetted and sinus tract was explored. On exploration a thin rim of bone lining a hollow cavity beneath 36 was revealed. The hollow cavity was curetted and the fragments of the excised tissue were sent for histopathological examination.

The patient's previous medical history was non contributory except that he was diabetic since last three years and was on oral anti-diabetic drugs. His blood sugar levels were under control. Routine haematological investigations were within normal limits. There was no history of immune deficiency or underlying disease. Physical examination revealed vital signs within normal limits. The laboratory results were unremarkable and the first hour erythrocyte sedimentation rate (ESR) was 12mm. Patient was a cigarette smoker with the frequency of eight cigarettes per day for the last ten years and was also, an occasional alcoholic.

HISTOPATHOLOGY

Sections from the excised tissue revealed numerous colonies of actinomycotic organisms surrounded by a collection of polymorphonuclear leukocytes (Fig. 4). The colonies consisting of club shaped filaments showed rosette pattern (on Haematoxylin and Eosin staining) with basophilic central core and eosinophilic peripheral portion. Few fragments of exfoliating necrotic bone were also evident. Gram's stain revealed a virtually pure population of Gram positive, predominantly filamentous organisms morphologically resembling members of the actinomyces species. Methanimine silver staining also demonstrated the colonies of actinomycosis.

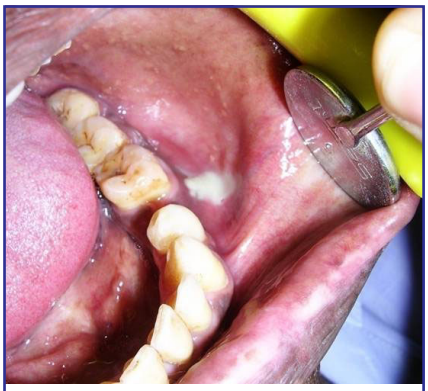

Fig. 1: Single pus discharging sinus in relation to 35 & 36 area with frank pus discharge

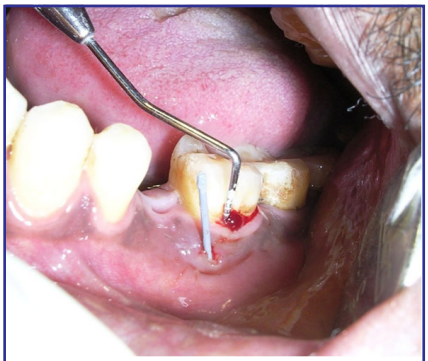

Fig.2: Picture depicting sinus opening and pocket depth in relation to 36

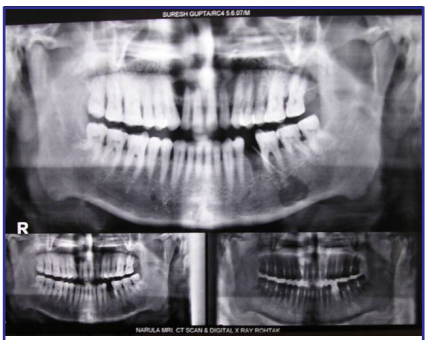

Fig.3 Panoramic radiograph showing lower extent of lesion

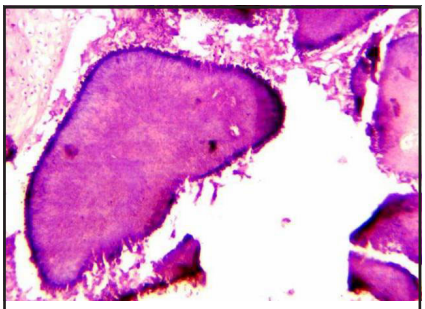

Fig.4: Hematoxylin and Eosin staining (40X) shows actinomycotic colony exhibiting club shaped filaments arranged in a radiating rosette pattern

The diagnosis of actinomycosis is achieved ideally by culture, but less than 50% of cases are positive because of the overgrowth of associated bacteria, prior antibiotic therapy or improper anaerobic media conditions. Due to lack of positive culture results, a strong presumptive diagnosis can be obtained through demonstration of the typical colonies in lesional biopsy material.

In our case the histological appearance of the biopsied material was consistent with those of chronic osteomyelitis in association with infection by actinomyces like organisms.

Periapical radiolucency in relation to the non vital upper right lateral incisor was also seen on the panoramic radiograph. To rule out multiple lesions caused by the actinomyces, a surgery was planned to excise periapical pathology in relation to 12 by raising the mucoperiosteal flap and excised tissue was sent for histopathological examination. The report revealed an inflammatory response with presence of inflammatory cells and absence of actinomycotic colonies.

Treatment with oral Amoxicillin with Clavulanic acid, 625mg, three times a day was initiated. Uneventful healing took place and pus discharge stopped by 7th day with no sign of recurrence clinically (Fig.5). Antibiotic therapy was continued for 3 months with follow up after every month(6).The patient had been asymptomatic for a period of eleven months following completion of antibiotic therapy. Panoramic radiograph taken six months after discontinuation of

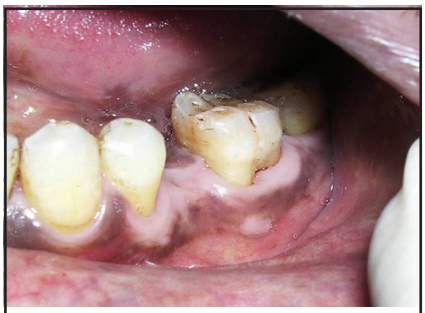

Fig.5: Uneventful healing and no recurrence clinically

treatment was interpreted as indicative of an increase in new bone formation (Fig.6).

DISCUSSION

Osteomyelitis due to actinomyces has been reported infrequently in adults. Bone involvement occurred in 1 to 15 percent of those series reviewed by Lewis and associates (6). Haematogenous spread of actinomyces with intra osseous granuloma formation and minimal sub periosteal bone reaction has been reported by Gholamreza R et al (7). However, the involvement of the mandible in our patient was probably by direct extension from either a periodontal tissue or a soft tissue focus.

Actinomycosis has been referred to as the chameleon of head and neck pathology because of its varied clinical picture which can resemble various pathologies ranging from benign infection to metastatic tumor (8). Cervicofacial actinomycosis should be included in the differential diagnosis of any soft tissue swelling in the head and neck region, particularly if malignancy or a granulomatous disease is suspected. The diagnosis of actinomycotic osteomyelitis often is overlooked because of this entity's ability to mimic other conditions (9).Mandibular actinomycotic osteomyelitis also is underappreciated by many clinicians in their assessment of head and neck infections(10).Most cases are traced to an odontogenic source, with peri apical tooth abscess and post traumatic or surgical complication as key antecedent events(11). Hence a biopsy should be performed on any persistent periapical lesion with osteomyelitis, even though a chronic draining sinus or cervicofacial abscess does not exist.

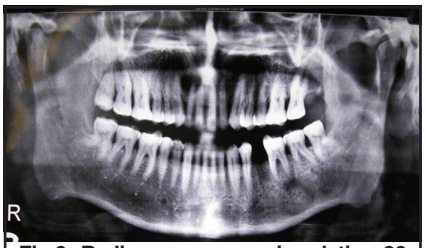

Fig.6: Radio opaque area in relation 36

Surgical treatment is also recommended in actinomycotic osteomyelitis cases in order to achieve two goals: removal of necrotic tissue and penetration of antibiotics into the colony of microorganisms which is inaccessible otherwise, either because of fibrous tissue or surrounding edematous tissue (12).

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