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CASE REPORT

ACTINOMYCOSIS 'AN UNLIKELY OFFENDER' - A CASE REPORT

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ABSTRACT:
Actinomycosis is a chronic, suppurative granulomatous infection caused byactinomycosis species, involving only the bone and the soft tissue or both. It is caused by a normal commensal developing opportunistic traits. On account of the rarity of the condition it is often misdiagnosed as a malignancy. In the present case the patient had repeated swellings in the mandibular region that was finally interpreted to be actinomycosis at a later date after thorough histopathological examination.
Key-words: Actinomycosis, Hoepple Splendore effect, Cervicofacial actinomycosis
Key Messages: In the diagnosis of chronic recurrent swellings of the mandible actinomycosis should also be considered regardless of its rarity.

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INTRODUCTION:

Actinomycosis is an infrequent, invasive bacterial disease recognized since a century. The species are filamentous, gram positive bacilli; a human commensal bacteria of the oropharynx, gastrointestinal tract and urogenital tract.[1] More than 30 species have been described Most cases are odontogenic in origin and occur predominantly in immunocompetent individuals. The microorganisms are generally of low pathogenicity and cause disease only in the setting of antecedent tissue injury8.Once infection is established the host mounts an intense inflammatory response and fibrosis may develop subsequently and spreads, frequently ignoring tissue planes.[2]

CASE HISTORY:

A 28 year old male patient was referred to the Department of Oral and Maxillofacial Pathology for evaluation of a swelling on the left side of the face, of one week duration associated with mild pain. The patient had a history of trauma 2 years back .Following the trauma the patient had developed a similar swelling accompanied by trismus in the same area.

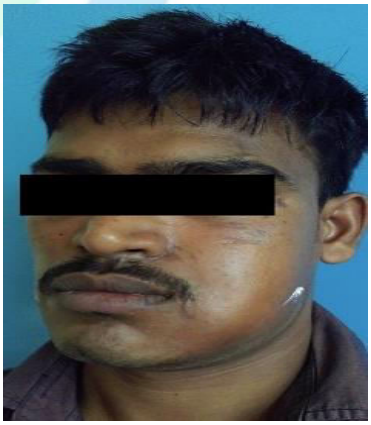

Figure 1: Clinical presentation.

Hospital records showed that intra-orally in the canine pre molar region there had been a lacerated wound that was subject to its debridement, surgery and subsequent medication. Incomplete record of visits to hospitals for recurrent swellings in the area was noted. This visit to our Dental OPD was with a complaint of an increase in size of the swelling over a period of one week

Extra oral examination revealed a diffuse swelling on the left side of the face extending from the

corner of the mouth to about 3 cm ahead of the ramus of the mandible, inferiorly it extended from the corner of the mouth to the lower border of the mandible. On palpation the swelling was tender, bony hard in consistency and there was a local rise in temperature. The submandibular lymph nodes were found to be enlarged, palpable and tender.
Intra oral examination revealed poor oral hygiene status. Vestibular tenderness was observed in relation to 34 and vestibular obliteration in the same area was also noted. Radiographic findings were non-contributory.

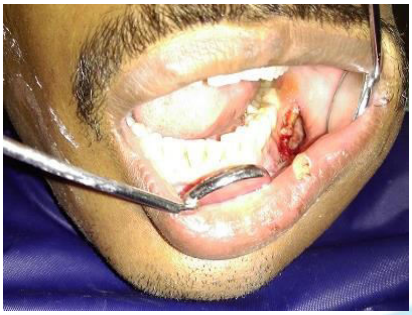

Figure 2: Intra oral vestibular obliteration.

The patient was admitted to the hospital and surgical exploration of the lesion was performed under general anaesthesia. A buccal sulcus incision was placed and surgical exploration was done till the lower border of the mandible. Multiple small black particles embedded in the muscle were seen and the tissue was found to be totally adherent to the bone. Multiple tissue fragments were sent for histopathological evaluation.
A biopsy specimen was submitted for histopathological evaluation. The biopsy revealed fibro muscular connective tissue with granuloma formation. Giant cell granulomas with colonies, in the centre morphologically consistent with actinomyces species were seen. PAS stain demonstrated colonies resembling sulphur granules that supported the H& E diagnosis of actinomycosis.

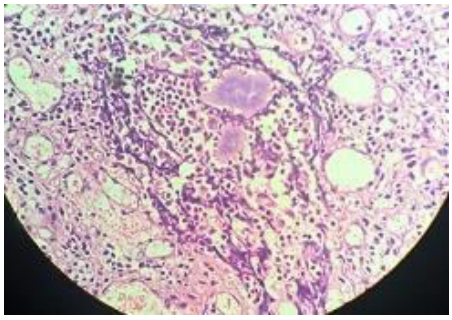

Figure 3: H&E stained section showing Hoepple Splendore effect

Gram staining further revealed gram positive interwined filamentous branching filaments consistent with the diagnosis.

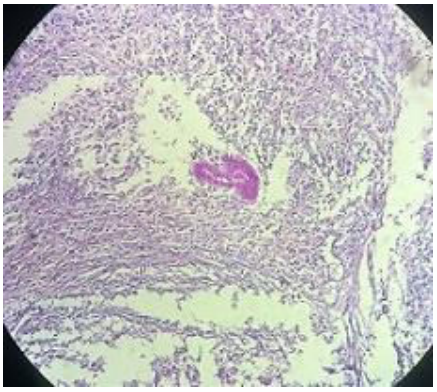

Figure 4: PAS stained section.

The typical picture of actinomycosis represented by granulation tissue in the outer zone with necrosis in the centre giving rise to the Hoepple Splendore effect was seen. The patient was prescribed Taxim, Metrogyl and Gentamycin and placed under observation, till his discharge three days hence. He was asked to continue with the medication and report for review every 4 months. A resolution of the swelling was observed at the first review.

DISCUSSION:
In the human species actinomyces are frequently part of the normal flora oforopharynx, gastrointestinal tract and female urogenital tract. They only cause infection in the presence of local predisposing factors6 like a complication of maxillofacial trauma, after surgical procedure and dental manipulation in a patient presenting poor oral hygiene or caries.[10] Cervicofacial actinomycosis is the most common form of this rare disease. These bacteria cannot penetrate healthy tissue and mucosal breakdown is a pre requisite for infection10. The infection usually presents as a chronic, suppurative, indurative mass, with discharging sinuses frequently located at the lower border of the mandible with or without lymphadenopathy.[10] Clinical diagnosis is often difficult due to non -specific findings. Differential diagnosis includes many diseases from various chronic diseases to tumours. Cervicofacial actinomyces could be associated with large abscesses and mandibular osteomyelitis with or without sinus tract and lead to distant organ dissemination1. Radiographic findings have little if any role in the diagnosis of the lesion. CT and MRI scans may show a non-specific involvement of skin and soft tissues but are useful to assess bone involvement1.It is important to emphasize that after

inoculation of actinomycetes or inadequate primary treatment, actinomycosis can recur years after the first inoculation. In the present case lack of proper initial histopathological diagnosis led to a delay in a definitive diagnosis after a 2 year period. However the reason why actinomycosis may manifest itself years after inoculation still remains unknown.[5] To obtain the definitive diagnosis incisional biopsy and histopathological analysis are necessary. Microscopically the granules manifest a cauliflower like shape at low magnification (×10).When pressed between the slide and the coverslip at higher magnification a clump of filamentous actinomycete microcolonies surrounded by polymorphonuclear neutrophils (PMNs) can be observed .Gram stain renders the microcolonies visible as Gram positive interwined branching filaments with radially arranged peripheral hyphae.[5] On histopathological evaluation of the present case the classic picture of actinomycotic colonies in granulomatous tissue was seen. Sulphur granules were revealed through PAS staining as well. Because of the diagnostic difficulties actinomyces is also known in literature as the great 'mimicker 'or when referring to cervicofacial actinomycosis as a great 'masquerader' of head and neck diseases5 Long term antibiotic treatment is advisable to prevent chances of relapse. Pencillin G is the drug of choice for treating infections caused by actinomycosis.

CONCLUSION:

Actinomycosis needs to be considered as a differential diagnosis in persistent or long term pathologies relating to the cervicofacial region. Most patients as in the present case give a history of maxillofacial trauma to the region. Clinical along with histopathological assessment is mandatory for early diagnosis. This can prevent the occurrence of possible complications through osseous and haematogenous spread and minimize aesthetic and functional damage.

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