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Research Paper

Actinomycotic Osteomyelitis of the Maxilla presenting in the form of Oro-Antral Fistula: a Case Report

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ABSTRACT *-Actinomycosis is a chronic granulomatous disease which is caused by Actinomyces species which may involve soft tissue , hard tissue and even both. The osteomyelitis of the maxilla arising as a result of Actinomyces species is relatively rare as compared to the mandible. These exist as commensals and cause infection when they gain entry into various tissue spaces where they cause extensive fibrosis and sclerosis as a result of the surrounding anaerobic environment . The mode of entry of the infection may be via the pulp or the periodontal ligament which may even cause the involvement of adjacent structures such as the pharynx , larynx , tonsils and even the paranasal sinuses .The diagnosis is often delayed and is dependent on the histopathological analysis .The management of actinomycotic osteomyelitis consists of surgical debridement of the necrotic tissue followed by antibiotic cover for a period of 3-6 months. Here , we present a case of a 55 year old male with actinomycotic osteomyelitis of the maxilla which presented clinically as an oro-antral fistula with suppurative features.*

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I. INTRODUCTION

Actinomycosis is a saprophytic infection which is characterized by granulomatous lesions that are caused by resident oral microbiota, Actinomyceteceae. They are slow growing , filamentous bacteria that resemble fungi . Actinomyces species are gram positive ,non acid fast , anaerobic and microaerophilic bacteria. Most of the species isolated from the lesions have been identified as A.naeslundii , A.israelli, A.viscosus and A.meyeri. [1] It may involve the soft tissue, hard tissue (osteomyelitis) or both . It can present in an acute , subacute or a chronic form.The incidence of Actinomyces infection in the mandible is 53.6 % followed by cheek (16.4 %) , chin (13.3%) , maxilla (5.7 %) and the TMJ (0.3%).[1] The infection spreads in a contiguous manner , frequently ignoring the tissue planes or the investing tissues or organs , producing sinus tracts . [2]Actinomycotic osteomyelitis of the maxilla is relatively rare as compared to the mandible probably because of better circulation which provides an increased oxygen supply .[2] The incidence of primary actinomycosis of the maxilla arising as a result of the contiguous involvement of the paranasal sinuses with the formation of oroantral fistula and palatal perforations is rare. [3,4,5,6,7]. This case depicts actinomycotic osteomyelitis of the maxilla that presented as oroantral fistula with suppurative features in a 55 year old male .

II. CASE REPORT

A 55 year old male presented with a chief complaint of unhealed tooth socket and opening in the area in right upper back tooth region since 3 months . The case gave a history of extraction of upper right tooth 1 and a half year back . The patient then gives a history of a painful swelling in the same area after 3 months .He was put on antibiotics and pain relief medication by the local doctor in the area .On no relief , the patient again reported to the dentist who performed a surgery for the drainage of pus for which the patient has no records. He also complains of fluid discharge through the right nostril on consumption of water orally .

An intra oral examination shows an irregular necrotic defect in the maxilla with antral communication , forming an oroantral fistula in the region of 14, 15. There was a purulent discharge from the adjacent bone area.

The defect was non – tender on palpation and the bone was soft to firm in consistency (Figure 1 and 2). 11 was tender on palpation and showed Grade II mobility.

Figure 1 – Clinical photograph of the communication

Figure 2 – Clinical picture of the oroantral fistula

The patient was advised a CT face , routine hematological examination and bone biopsy. The CT revealed a destructive lesion that involved the right maxilla extending upto the right infra orbital margin . (Figure 3). The lymph nodes were non palpable and there was no evidence of lymphadenopathy.

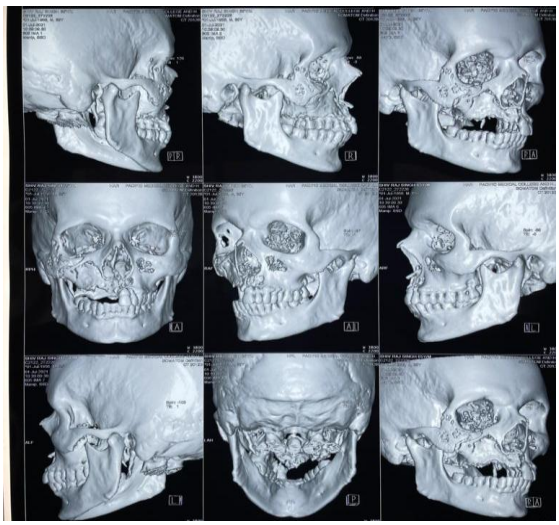

Figure 3 – CT Face showing erosion of bone.

The incisional biopsy taken from the bone revealed a stratified squamous epithelium with large clumps of basophilic bacterial colonies having a sun-ray appearance which is a characteristic feature of actinomycoticinfection. The routine hematological examination was normal except for an increased neutrophil count and elevated Erythrocyte sedimentation rate (ESR) .The final diagnosis of Actinomycotic Osteomyelitis was reached.

The patient was operated under general anesthesia and the procedure consisted of debridement of the infected area with the removal of the bony sequestra through a Weber –Ferguson approach (Figure 4). 11 was also extracted and the tissue was sent for histopathological examination.(Figure 5). The patient was kept under penicillin IV post operatively for a period of 1 month and Doxycycline 100 mg twice a day (bid) for the following 2 months.

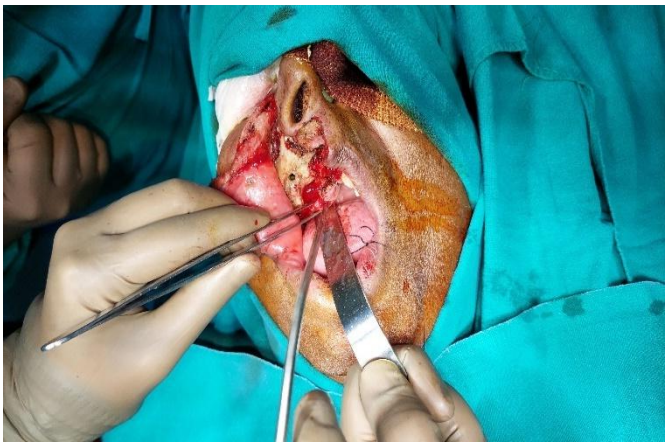

Figure 4 – Weber Ferguson approach

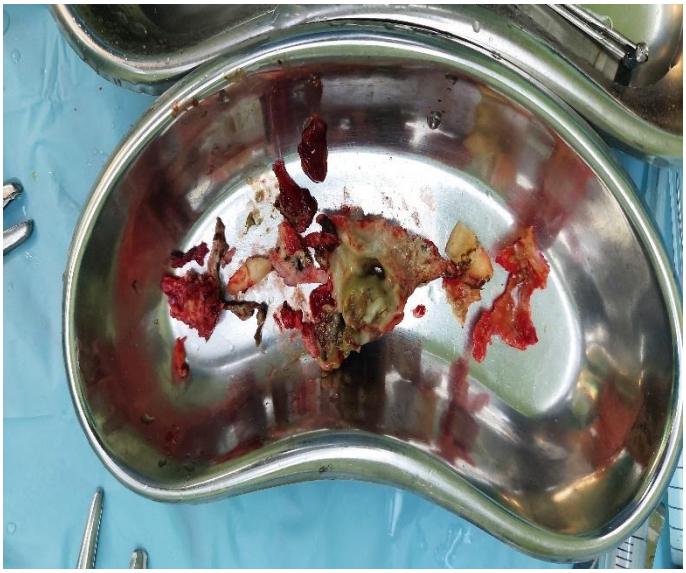

Figure 5 – The surgical specimen

The surgical specimen was histologically evaluated and showed extensive sclerosis of bone with prominent resting and reversal lines . There was evidence of extensive fibrosis with resolving granulomas.
The patient was recalled after one month and showed improvement with near complete resolution of the lesion . (Figure 6). The patient is still on regular follow up and till now there is no recurrence .

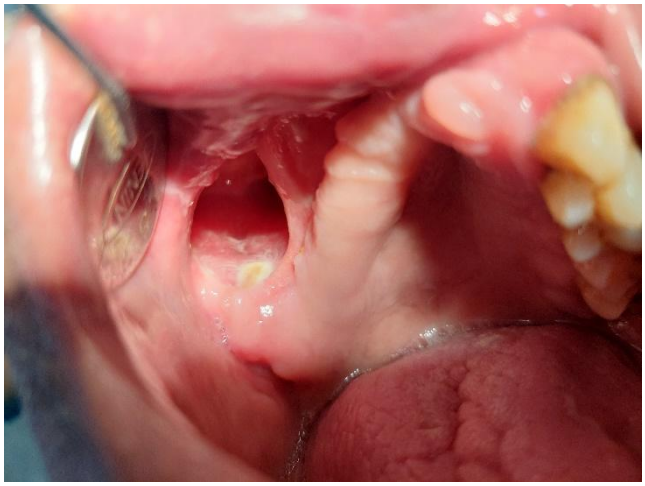

Figure 6

III. DISCUSSION

Actinomycosis in the maxilla accounts for 0.5 – 9 % of all head and neck cases. Although the exact underlying pathophysiology of actinomycotic osteomyelitis is unclear , it is suggested that the inflammatory process begins with the disruption of the normal composition of the oral microbiota and thereafter, the chronic inflammation leads to localized pathological changes in the bone . [8]

Marx et al has described a more refractory form of chronic diffuse sclerosing osteomyelitis produced by Actinomyces together with other organisms such as Arachnia and Eikenella. It has 2:1 predilection for women and an average course of nearly 5 years before diagnosis .[9].The primary osteomyelitis of maxilla with the involvement of maxillary sinus can arise as a result of adjacent oral soft tissue infection due to periapical or periodontal pathology. The underlying criteria for the development of endogenous disease is the transport of the pathogens into deeper tissue layers with anaerobic environment. The chronic purulent infection with unhealed sockets in this case show that the penetration site of these organisms into the deeper tissues was from gingiva, periodontal disease or chronic periapical abscess. [6,7] The infection presents itself as granulomatous inflammatory response with central suppurative necrosis that consist of aggregates of bacterial filaments that are surrounded by neutrophils .

Experiments have shown that pure cultures of Actinomyces produce acute suppurative infections and mixed infections produce lesions with A.actinomycetemcomitans that evade direct contact with antibiotics and leucocytes within the sulfur granules.[9] In the long standing cases , the fibrosis can lead to end level sclerosis which in turn can lead to therapeutic failure as poor vascularity inhibits the penetration of antibiotics. The treatment should be vigorous with complete debridement of the necrotic tissue and resection of the sequestrated bone until healthy and viable tissue is exposed .The drug of choice is penicillin with the course ranging from 3 to 12 months and erythromycin ,tetracyclines are the effective alternatives .

The diagnosis is often delayed because of different presentations and they are cultured in less than 50 % of the cases .[1] A clinical diagnosis may often be difficult and various problems are associated with bacteriological culture making histopathological analysis a necessity.The nucleic acid probes and polymerase chain reaction (PCR) methods have been deployed for rapid and accurate identification but are highly expensive .[10]

IV. CONCLUSION

Oral mucous membrane is often the penetration site of Actinomyces into deeper tissues. Definitive diagnosis based on cultures is not often positive but histopathology still remains the most reliable diagnostic aid. It is a potentially benign and completely curable disease with an early diagnosis and adequate therapy is absolutely essential so that there is minimal functional and esthetic damage.

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