



PRIMARY CUTANEOUS ACTINOMYCOSIS OF CHIN WITH OSSEOUS METAPLASIA – A CASE REPORT

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ABSTRACT

Actinomycosis is a granulomatous suppurative bacterial disease caused by anaerobic actinomyces, Primary cutaneous actinomycosis is a rare entity usually associated with external trauma and/or local ischemia and the diagnosis requires a high index of clinical suspicion. Anaerobic cultures may be negative despite repeated attempts. Microscopic examination reveals the diagnosis in the majority of cases. We report a case of a primary cutaneous actinomycosis affecting the chin in a 22-year-old man .To the best of our knowledge, this is the first case of primary cutaneous actinomycosis affecting the chin with osseous metaplasia.

Key Words : Primary ,Cutaneous, Actinomycosis, Osseous Metaplasia

Introduction

Actinomycosis is a chronic and suppurative infection caused by an endogenous gram-positive bacterium, *Actinomyces israelii* which presents primarily with the cervico-facial, thoracic, abdominal or pelvic form¹. Primary disease of the skin is uncommon and has an association with trauma and bites. The main differential diagnosis is nocardiosis; the distinction can be made by the histopathological examination aided by special stains².

Case Summary

A 22-year-old farmer visited our hospital with complaints of reddish elevated itchy soft lesions over the right side of the chin since three months. which gradually increased and transformed into multiple elevated cystic lesions . He consulted a local doctor with no response to treatment and lesions discharged serous fluid.

Local examination of right side of chin showed multiple reddish elevated cystic lesions with discharging sinuses extending over an area 6×5 cm associated with diffuse thickening of the skin and subcutaneous tissue (Fig 1 & 2).



Fig 1: Ill-defined, 5x5 cm (approx), erythematous ,indurated plaque with overlying vesicles, nodules



Fig 2: Erythematous papules & nodules present over mucosal aspect of lower lip.

His systemic examination, hematological and biochemical parameters were normal. X-ray of the mandible did not show any evidence of osteomyelitis. HIV-ELISA was non-reactive. X-ray chest showed no involvement of lungs or vertebra. USG of the involved area showed a 4.5x3.9x2.8cms soft tissue lesion over the right mandibular region in subcutaneous region with moderate vascularity and few hyperechoic foci (?calcification) suggestive of chronic granuloma or neoplasm. Fine-needle-aspiration cytology revealed nonspecific acute and chronic inflammation. The acid-fast-stained smear for *Mycobacterium tuberculosis* was negative. Repeated microbiological cultures of a skin biopsy specimen for both aerobic and anaerobic bacteria failed to reveal any growth. Culture for *M. tuberculosis* and fungi was negative. A lesional biopsy showed multiple microabscesses in the dermis with the presence of abundant lymphocytes, plasma cells, neutrophils, and macrophages with one abscess showing an actinomycotic colony (Fig 3).

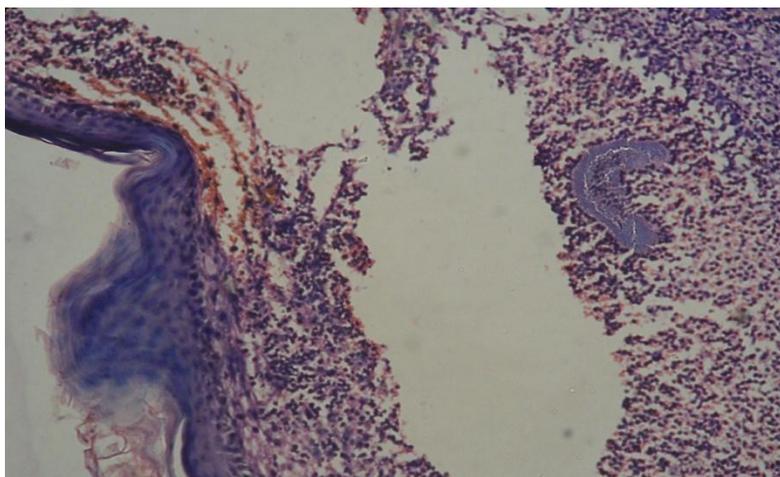


Fig 3: Microphotograph showing the polymorphous inflammatory infiltrate with an actinomycotic colony (H&E, 40 X)

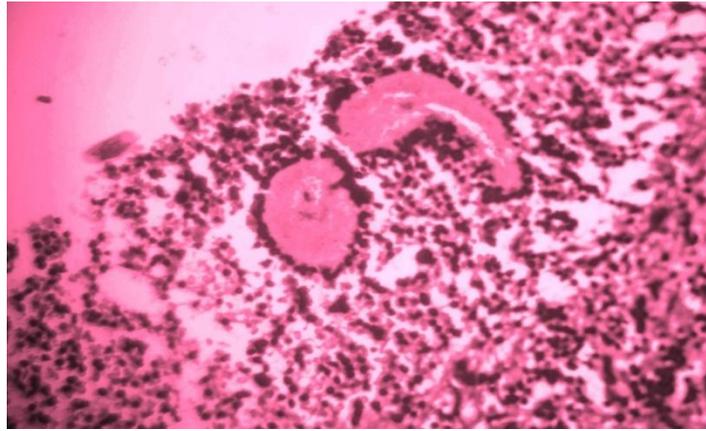


Fig 4 : Microphotograph showing actinomycotic colony admixed with inflammatory infiltrate (H & E,100X)

A focus of osseous metaplasia was noted .Gram staining revealed numerous, Gram-positive, branching, filamentous, rodlike organisms intertwined in the granules Gram staining and staining with PAS confirmed actinomycotic colonies .We rendered the final diagnosis of primary cutaneous actinomycosis with osseous metaplasia.

Discussion

Actinomyces israelii is a normal inhabitant of the oral cavity. Infections are considered to be endogenous and the organism becomes pathogenic in the presence of devitalized tissues with reduced oxygen tension. Thus, actinomycosis may develop in the jaw and neck after an infection or dental surgery or in lung or gut when superimposed on an antecedent disorder that provides a favorable environment for its growth (e.g., lung abscess, ulcero-inflammatory disease of the gut)². Primary cutaneous actinomycosis occurs on exposed skin through implantation. Although it occurs worldwide, it has a higher incidence in farm workers of rural tropical regions, possibly related to poor oral hygiene and limited use of antibiotics effective against actinomycetes in these areas. Actinomycosis has a peak incidence in the middle decade with three-fold higher incidence in males ³.In our case, the 22 year old male patient was a farmer with no history of trauma or operation in the nasal and oral cavities so the exact pathogenesis remains unclear. Grossly, it presents as intense suppuration with abscess and sinus formation discharging yellow sulphur granules. In our case, it was the lesion having typical presentation with abscess on the jaw . Histology is diagnostic, characteristically shows colonies (sulphur granules) formed by

tangled mass of filaments surrounded by radiating, sometimes terminally clubbed, organisms¹. Special stains and the shape of the microorganisms are important because sulfur granules are not unique to actinomycosis. They can be found in nocardiosis, chromomycosis, eumycetoma and botryomycosis⁴. Moreover the management of the patient differs depending upon the organism. The treatment of actinomycosis requires administration of parenteral or oral penicillin for at least 6 weeks whereas nocardiosis responds to anti-fungal treatment. In our case, sulfur granules consisted of filamentous, rod-like organisms that were positive for Gram and negative for Ziehl Neelsen stain. In nocardiosis, they are usually Ziehl Neelsen positive. Gram stain is negative for chromomycosis and eumycetome. In botryomycosis, microorganisms are non-filamentous bacteria. Though the bacterial culture for *Actinomyces* is essential, it needs strict anaerobic conditions and the positive rate is reported as low as 35%^{2, 4}. We carried out bacterial cultures three times but none were positive. However, the histopathological examination including special stains revealed sulfur granules that were consistent with actinomycosis which established the diagnosis.

Cutaneous actinomycosis is treated surgically followed by antibiotics, the most widely used of which are long-term penicillin, tetracyclin and erythromycin, and treatment period is variable, since the best dosage is unknown. In our case, characterized by a single, exclusively cutaneous lesion, brief treatment was sufficient as recently reported in the literature.

From this case we conclude that one should be very vigilant when such infections are seen in patients like farmers who are more prone for vegetative penetrating injury. An awareness of the full spectrum of the disease will expedite its early diagnosis and treatment and will minimize the unnecessary surgical interventions, morbidity and mortality.³

References

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