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Case Report

Orthopaedics Section

Actinomycosis Calcaneum- A Rare Case Report

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ABSTRACT

Actinomyces is anaerobic, gram-positive bacilli that superficially resemble fungi and give rise to suppurative and granulomatous infection. The organisms are commensals to human body but under suitable conditions can involve any organ system. Cervicofacial actinomycosis is the most common presentation while actinomycosis of extremity especially calcaneum is very rare. We report a rare case of 40-year-old woman who presented with chief complaints of pain and swelling of the right foot for one year. She was diagnosed with isolated calcaneal actinomycosis and was managed by antibiotics, curettage of calcaneum and triple arthrodesis.

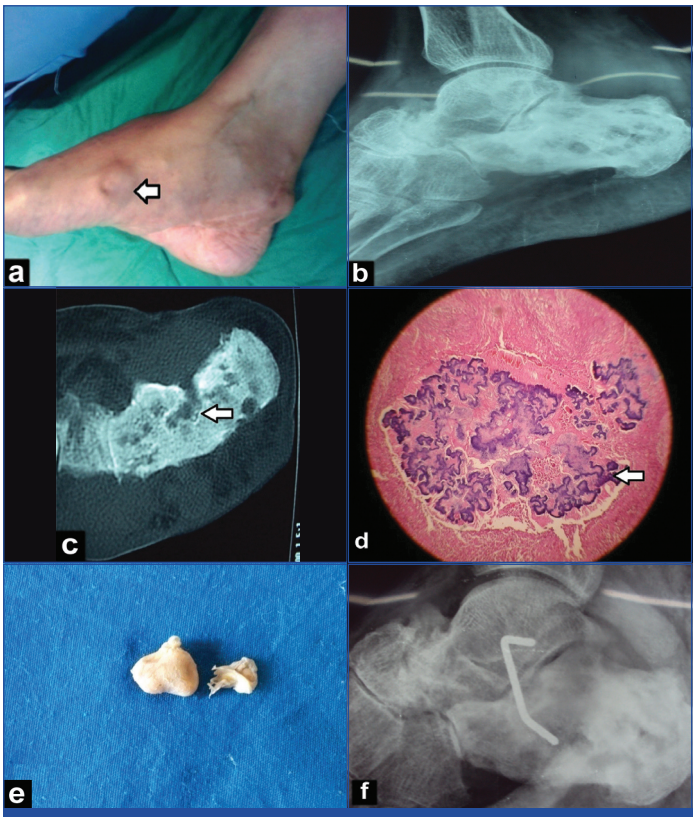
Keywords: *Actinomyces*, Calcaneal osteomyelitis, Triple arthrodesis

CASE REPORT

A 40-year-old woman from rural Maharashtra presented with chief complaints of pain and swelling of the right foot for one year. There was no history suggestive of recent or past trauma, fever, loss of weight, anorexia, other joint involvement, underlying systemic disease or being immunocompromised. Consent for examination, investigation and subsequent management was taken from the patient. Clinical examination revealed a 2x2 cm swelling on the dorsolateral aspect and similar two swellings medially in hindfoot region [Table/Fig-1a]. There was mild local rise of temperature and a slight tenderness at subtalar joint. There was no regional lymphadenopathy. Ankle joint movements were normal while subtalar joint movements were painful and restricted.

Radiography revealed soft tissue swelling around the ankle, osteopenia of underlying bones, sclerosis of calcaneum along with multiple osteolytic lesions in calcaneum [Table/Fig-1b]. Additional findings like narrowing of calcaneocuboid joint space were revealed on subsequent CT scan [Table/Fig-1c]. The possible radiological differential diagnosis entertained were chronic pyogenic osteomyelitis of calcaneum, Tuberculosis calcaneum and mycetoma (bacterial/fungal). Aspirate from the medial hindfoot swelling was subjected to histopathology and culture. On H&E stain, basophilic *Actinomyces* colonies were seen in the central haematoxylin-stained area surrounded by fibrocollagenous tissue, dense infiltrate of neutrophils, lymphocytes, plasma cells and occasional giant cells [Table/Fig-1d]. Simultaneously part of aspirate sent for anaerobic culture revealed *A israelii* as the causative organism. Culture of the aspirate on blood agar revealed *Staph aureus* as copathogen.

Tissue specimen removed at surgery comprised of necrotic bone debris and sulphur granules of variable size. Analysis of tissue specimen removed at surgery which consisted of necrotic bone debris and sulfur granules of varying size (2 mm to 2.5 cm) confirmed the diagnosis [Table/Fig-1e]. Patient was managed by thorough debridement of calcaneum and triple arthrodesis (Ollier's approach) [Table/Fig-1f]. Postoperatively patient was managed by below knee cast for weeks. The response to penicillin therapy, curettage and triple arthrodesis was satisfactory in the patient. Regular follow ups were conducted at monthly interval. Patient was disease free at one year follow up without any reactivation or recurrence.



[Table/Fig-1]: a) Swelling on the medial aspect of foot and heel; b) Preoperative X-ray of calcaneum (lateral view) showing ostelytic lesions and sclerosis; c) Axial CT scan showing calcaneum with multiple osteolytic lesions (arrow); d) H&E stain of sample showing basophilic *actinomyces* colonies in the central haematoxylin-stained area (arrow) and dense inflammatory infiltrate composed of neutrophils, lymphocytes, plasma cells; e) Specimen of the granules taken out during the surgery (2 mm-2.5 cm); f) Postoperative X-ray showing curettage and triple arthrodesis.

DISCUSSION

Actinomycosis can present in various clinical forms e.g., cervicofacial, thoracoabdominal and pelvic, the latter being common in females. *Actinomyces* is normal flora of oral cavity and gastrointestinal tract. Primary actinomycosis of an extremity is very uncommon because of the endogenous habitat of the organism [1]. Haematogenous spread from some other primary site or direct extension following a mucosal breech are the possible mechanisms [1]. Males are more commonly affected than females (3:1) [1].

Few studies from literature have reported actinomycosis of the metacarpal bone [2,3], forefoot [1,4], lower leg [5] and calcaneum [6]. Lack of information among clinicians and population regarding disease aetiology, presentation and progression frequently leads to a delayed diagnosis and a late initiation of treatment [7].

Primary actinomycosis, mycetoma (actinomycetoma and eumycetoma) and chronic pyogenic osteomyelitis of calcaneum superficially resemble each other in their clinical presentation. But difference in causative organisms and sensitivity to different chemotherapeutic agents makes them discreet clinical entities. Causative organisms of actinomycosis are *A israelii* and related anerobic or facultative bacteria, while, actinomycetoma and Eumycetoma are caused by *Nocardia* spp. and Fungi respectively. *S. aureus* is the most common causative organism for pyogenic chronic osteomyelitis [8,9].

Primary actinomycosis involving extremity is very rare. Reiner SL et al., reported 36 cases of primary actinomycosis of extremity worldwide. Two more cases were reported with last case being reported in 1996 [6,10-12]. The present case of primary actinomycosis calcaneum being reported from India is one more addition to it. On the contrary mycetoma foot is quite common in India [13,14]; while incidence of 11% has been noted for chronic pyogenic osteomyelitis of calcaneum [15].

Mycetoma foot and ankle is worldwide in distribution with higher prevalence in geographic area between 15°S and 30°N also known as the mycetoma belt. But no racial and geographical predilection has been noted for actinomycosis calcaneum caused by *A israeli.* The latter affects people with middle age who have *A israeli* as part of their flora [6,13,14] The present case was a 40-year-old female from rural Maharashtra, India.

Pyogenic osteomyelitis of calcaneum may present with history of hindfoot surgeries, scars and signs of infection like reddening of the skin, hindfoot pain, oedema and toe walking [16]. Actinomycosis of extremity may present with history of trauma to extremity, immunocompromise or predisposing factors like rural background, poverty, malnutrition, poor oral hygiene, agricultural occupation and bare foot walking [1]. In case of actinomycosis of foot or calcaneum there is swelling, induration, multiple sinuses and exudate containing granules. Chronic infection and progressive destruction are hallmark of the disease. As pain and inflammatory signs are absent, so the individual keeps walking on a diseased limb leading to further destruction. Mycetoma is a relatively painless condition with possibility of a high incidence of secondary bacterial infection, deformities and recurrences of the affected foot.

In present case, calcaneum was affected by actinomycosis, a rarely reported presentation in literature. Patient had swelling on the dorsolateral aspect and similar two swellings medially in hindfoot region. There was mild local rise of temperature and a slight tenderness at subtalar joint. There was no regional lymphadenopathy. In another similar case presentation of calcaneal actinomycosis in the form of hindfoot swelling, moderately discoloured hyperpigmented skin on the lateral aspect of right heel, with two non draining sinuses and painful subtalar movements was noted [6]. Histopathology and culture is cornerstone to establish diagnosis. Material exuded in discharge from sinus or core needle biopsy from the site of pathology, is subjected to histopathology and culture to arrive at a diagnosis [6]. Colour of granules is helpful as white granules indicate actinomycosis, white to yellow granules actinomycotic mycetoma, while mycotic mycetomas have black granules. Gram-positive branching bacilli in tissue exudates or on culture confirm the diagnosis [17]. Actinomycotic infection can occur in isolation but most of the times it is a polymicrobial infection with multiple organisms being cultured in as many as 99% of cases of actinomycosis [2]. If any surgical debridement is planned, the tissue removed at the time of surgery can be re-examined by histopathology and culture to reach a conclusion [6]. Achieving a specific diagnosis is important as actinomycosis and Mycetoma are sensitive to different antibiotic treatments.

Molecular identification by 16S PCR aids in *Actinomyces* species identification [16]. In present study also it was a mixed infection with methicillin sensitive *S aureus and A israelii* being isolated on culture.

On pathological examination of specimen, bones are found riddled with spheroidal defects with sclerosis and periosteal reaction at periphery. In advanced stage, gross destruction and disorganisation of underlying bones is the norm. Radiography reflects the pathological changes showing circular lytic lesions and a sclerotic margin. Possible radiological differential diagnosis are multiple myeloma (punched out lesions without sclerosis) and fungal osteomyelitis ("fronts of resorption" and penetrating spicules) [18]. Osteopenia of underlying bones and multiple lytic lesions in calcaneum were noted in present study.

Since, infected and indurated tissues are avascular, surgical debridement along with high dose of antibiotics for prolonged period (6-12 months) are mandatory [19]. Penicillins are drug of choice for actinomycosis followed by other antibiotics like erythromycin, tetracycline, clindamycin, cephalosporins, meropenem, and chloramphenicol [20], while *Nocardia* are known to respond to Trimethoprim-sulfamethoxazole, amikacin and linezolid.

Antifungal agents like ketoconazole, miconazole and amphotericin B are used to treat fungal mycosis. Frequent co-infections with other microbes make combination therapy advisable.

In actinomycosis calcaneum, appropriate antibiotics and surgical management in the form of curettage of lesions or partial/complete calcanectomy is opted; depending upon the extent of disease. Limited disease in calcaneum can be managed by antibiotics, local curettage and debridement of the lesion while, large extent of disease with multiple sinuses is managed by partial/total calcanectomy. Literature findings mention partial and total calcanectomy as a mode of treatment for chronic osteomyelitis of calcaneum with non healing ulcer and refractory chronic osteomyelitis [21]. Surgical procedure in the form of debridement, curettage and triple arthrodesis was used in present study while another study from literature has reported management with total calcanectomy [6].

CONCLUSION

This case report highlights the importance of actinomycosis as a rare but important cause of chronic osteomyelitis of calcaneum. The diagnosis is likely to be missed for more common conditions with sinuses e.g., chronic bacterial and granulomatous conditions like tuberculosis. A close interaction between clinician, pathologist and microbiologist can help achieve accurate diagnosis.

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