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# CASE REPORT

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## Actinomycosis of the calcaneus – a case report

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### Introduction

Actinomycosis, discovered in cattle, has long been controversial, and was classified as a fungus because of its branching character. It has a slender, beaded, 0.3 to 0.5  $\mu\text{m}$  wide and 2 to 30  $\mu\text{m}$  long branching appearance.<sup>1</sup> The 'hyphae', however, are approximately one-fifth the diameter of known fungi. It was subsequently classified as a branching bacterium.<sup>1</sup>

Classically thought as the agent of bovine 'lumpy jaw', it has a very different skeletal presentation in humans.<sup>2</sup> One peculiarity of actinomycosis is the formation of masses known as 'sulphur granules'.<sup>3</sup> Such sulphur granules can be produced by *Nocardia*, *Actinomadura*, *Streptomyces*, and possibly as a botryomycosis secondary to *Staphylococcus*, as well as *Actinomyces israelii*, which typically causes actinomycosis.<sup>3</sup> Actinomycetes are prominent among the normal flora of the oral cavity. They are not virulent and require a break in the mucous membranes to invade the body.

Actinomycosis affecting the lower extremity is rare, and as a cause of calcaneal osteomyelitis, has not been reported. It is a subacute to chronic bacterial infection caused by filamentous, Gram-positive, anaerobic to microaerophilic bacteria that are not acid fast. The principal agent for human infections is *Actinomyces israelii*.<sup>4</sup> Typically, there is suppurative and granulomatous inflammation accompanied by the formation of multiple bone abscesses and sinus tracts that may discharge. In isolated cases of actinomycosis, the most common clinical forms are cervico-facial ('lumpy jaw'), thoracic, abdominal, and pelvic in women. Orthopaedic and dermatological manifestations predominate in disseminated actinomycosis.<sup>4</sup> Actinomycosis is a polymicrobial infection and requires the presence of companion bacteria which act as copathogens to enhance the invasiveness of actinomycetes.<sup>4</sup>

We report a rare case of isolated right calcaneal actinomycosis managed surgically by total calcaneectomy.

### Case report

A 32-year-old black male, teacher, presented with a 15-year history of chronic right heel pain, with intermittent episodes of swelling, predominantly in the lateral sub-fibular region, that improves by the extrusion of yellow pus from two sinuses. These episodes occurred approximately four times a year, with relative quiescence of symptoms in the intervening periods.

During the flare-ups, and for as long as he could remember, he saw several general practitioners who managed him with a range of antibiotics and analgesics. However, the pain was becoming unbearable, affecting his gait as well as performance of occupational and sporting activities, and significant night pain had been present for the last six months. He presented for his first consultation in a wheelchair.

Despite the chronic course of the illness, the young man looked well nourished, with a BMI of 31, no generalised lymphadenopathy, and no other foci of infection, apart from several dental caries of his molars in the lower jaw.

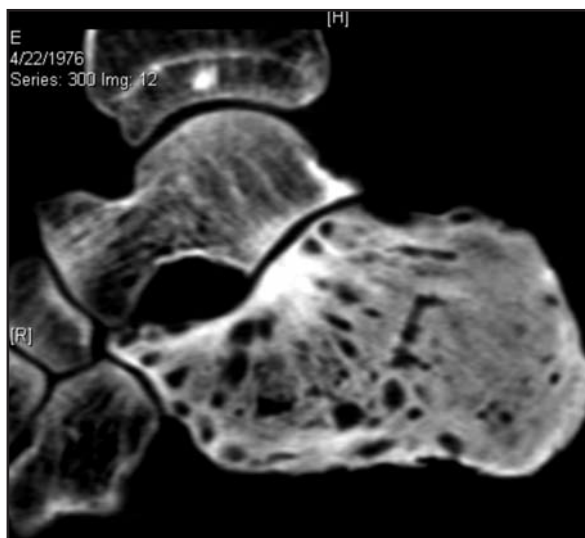
Examination of the right foot revealed moderately discoloured hyperpigmented skin on the lateral aspect of the heel, with two sinuses 2 cm below the tip of the lateral malleolus. These were not draining.

There was generalised hindfoot swelling with indurated soft tissue. The heel pad was healthy and the vascularity to the foot was normal.

Ankle movements were normal, but there was severe pain with movements of the subtalar joint. The ipsilateral calf was wasted.

The full blood count revealed a normocytic hypochromic anaemia, white cell count of 8.3 with a predominance of monocytes. The erythrocyte sedimentation rate was 48 mm/hr, the C Reactive protein was 6 mg/dl. Plain radiographs showed multiple spheroidal cystic lesions with reactive sclerosis throughout the calcaneus. This was confirmed on CT scan (*Figure 1*). The technetium bone scan showed increased uptake in the right calcaneus only.

Three core biopsies were taken under general anaesthesia and sent for microscopy, culture, fungal stains and histology. Microbiological studies were non-contributory, mycobacteria and fungi were not demonstrated on the specimens, while the histology demonstrated chronic granulomatous inflammatory infiltrates around the infected bone. The patient was started on antituberculous treatment empirically. The patient was still experiencing significant pain, inability to weightbear on the affected side and weight loss of 3 kg in two months.



**Figure 1:** Lateral CT scan of the calcaneus showing numerous spherical lesions with marginal sclerosis involving the whole calcaneus. Coalescence of lesions is seen



**Figure 2:** Post-operative appearance of the foot shows normal contour of the heel, a healed sinus and good dorsiflexion

The patient underwent total calcaneotomy two months later. After removal the calcaneus was split and examined. The cut surface showed numerous small spheroidal cavities, with a honeycomb appearance and multiple pockets of pus. The bone was very sclerotic.

The patient made good recovery. The wound healed well. Ankle motion ranged from 20° dorsiflexion to 30° of plantar flexion. The outline of the heel pad was maintained (*Figure 2*).

Culture reports revealed *Actinomyces israelii*. Histology showed bony trabeculae, with intervening granulation tissue and microabscesses. Colonies of organisms were seen in keeping with actinomycosis. Antituberculous medication was stopped and oral antibiotic treatment with penicillin was commenced for six weeks. At 4-month follow-up the patient was fully weightbearing with a heel cup and orthopaedic boots.

## Discussion

Actinomycosis originally described as a fungus is now considered a branching bacterium.<sup>1</sup> Infection is most commonly caused by *Actinomyces israelii*, a Gram-positive rod colonising in the mouth. There are several other species that can cause infection. Poor oral hygiene and dental caries are suspected portals of entry for infection.

There is no racial or geographical predilection, and it affects middle-aged adults, but only those who have the organism as part of their flora. Actinomycosis is not found outside the body.<sup>2</sup>

Cervicofacial, thoracic, and abdominal actinomycosis are the three most common forms of the disease. Actinomycosis can occur anywhere but is least likely in the extremities.<sup>2</sup> Involvement of the bone is not common in man even in facial actinomycosis and is extensive only with dissemination or with a protracted course. This is in contrast with typical bovine actinomycosis ('lumpy jaws') in which the mandible becomes eroded and expanded. Brown,<sup>2</sup> in a large study of 181 patients with actinomycosis, found extremity involvement in eight cases, the lower limb being involved in five cases (2.8%) only. Blinkhorn,<sup>3</sup> in a large review of upper limb involvement, found nine cases of osteomyelitis of the hand due to actinomycosis. He stressed the association of hand infection due to punch injuries. Soft tissue infection subsequently involving bone by contiguous invasion occurred following a laceration while striking the adversary's teeth. The infection was due to *Actinomycosis israelii* in all cases.<sup>2,4</sup> The term 'punch actinomycosis' was coined by Burrows<sup>5</sup> to describe this injury.

Cope, in 1951,<sup>6</sup> updated the literature on spinal involvement. He recorded 66 cases of vertebral actinomycosis and concluded that infection arises by contiguous spread from the deep cervical region, mediastinum, or via retroperitoneal infiltration. The disease may be linked to one vertebral body, but more usually, several vertebrae are affected. The intervertebral discs, pedicles, transverse processes, and heads of the neighbouring ribs may also be involved. Radiographs may show a lattice network due to rarefaction and sclerosis. He emphasised that spinal actinomycosis must be considered when diagnosing any subacute or chronic spinal condition.

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Involvement of the calcaneum has not been described.

Clinically actinomycosis presents as a chronic localised inflammatory process, with fever and leucocytosis. The diagnosis is often difficult to make, as only one-third of patients have sinuses. Abscesses with thick, yellow-green pus are characteristic of actinomycosis, but hard grains or granules are infrequently seen in the exudate. Lack of calcification may make granules difficult to find.<sup>2</sup>

Pathologically, bones affected by actinomycosis are riddled with spheroid defects associated with periosteal reaction. Erosion occurs through trabecular and cortical bones and the infection stimulates a reactive bone margin with coalescence of individual bone lesions.<sup>1</sup> Brown<sup>2</sup> described 'loculation' with fibrous walls, with no visible necrosis. Parker<sup>7</sup> described multiple cavities filled with granulations.

Radiologically, the lesions are circular, numerous, with slightly sclerotic margins.

In the treatment, cultural confirmation of the organism is important. Special care is required in the handling of specimens for microbiological study, for accurate diagnosis. The diagnosis is based on finding granules (grains) with clubs and Gram-positive branching bacilli in tissue exudates or on culture.<sup>2</sup> Actinomycosis is difficult to culture. Prompt transport of the specimens to the microbiology laboratory is necessary for optimal isolation, preferable in an anaerobic medium.

A high index of suspicion is necessary, since the grains may be scarce. Furthermore, other bacteria may form granules and this may be confused with actinomycosis. Infections due to *Nocardia*, *Actinomadura*, some *Streptomyces* species, as well as *Botryomycosis* may also produce sulphur granules. The finding of a polymicrobial infection is not surprising since multiple organisms have been cultured in as many as 99% of cases of actinomycosis.<sup>3</sup> Gram Stain reveals Gram-positive microcolonies, intertwined branching filaments with coexisting companion bacteria which are Gram-positive and Gram-negative cocci and rods. Penicillin for 6-12 months is considered the drug of choice for actinomycosis.

Therapy consists of surgical debridement with prolonged antibiotic treatment. The treatment of chronic osteomyelitis of the calcaneus is difficult especially when the whole bone or a large part of it is involved. Neither local nor general antibiotic therapy seems to be effective, because sclerosis of the bone prevents antibiotic penetration to the infected areas. Saucerisation followed by skin grafting has limited application. Reinfection is common.<sup>8</sup>

Gaenslen's operation which implies exposure of the calcaneus through a midplantar incision, and longitudinal splitting of the bone and removal of all dead bone and granulation tissue, results in a fragile scar and a high recurrence rate.<sup>9</sup>

Calcanectomy has been preferred as a method to eradicate the chronic infection.<sup>8</sup>

Partial or total calcanectomy is a relatively rare orthopaedic procedure. The indications are mainly for chronic osteomyelitis of the calcaneum, chronic heel ulceration in diabetes, and severely comminuted fractures.<sup>8,9</sup> Crandall reviewed 31 cases, 20 partial and 11 total calcanectomies, and found good results in non-diabetics. Only one patient developed midtarsal subluxation. Martini<sup>13</sup> performed calcanectomy in 20 patients, nine of whom had haematogenous osteomyelitis. Only one was younger than 10 years, with the majority being adults. Eighteen patients were able to weightbear on the heel.

He described the removal of the calcaneus, partially or totally through a midline longitudinal incision above the heel, to the midplantar surface of the foot.

The spectrum of clinical and radiological appearances of calcaneal lesions of osteomyelitis may mimic a host of disorders. Although bone involvement in chronic and subacute pyogenic osteomyelitis is common especially in children, the permeative reaction associated with sclerosis can resemble several other conditions. The differential diagnosis should include tuberculosis, 'atypical' mycobacteria, fungal infections, sarcoidosis, and certain neoplasms.

Tuberculosis of the foot is an uncommon presentation of skeletal tuberculosis. Dhillon<sup>10</sup> found calcaneal involvement was the commonest site in 74 cases of foot and ankle tuberculosis. The commonest presentation was a cystic lesion with or without a sequestrum. Sinuses were seen commonly. Similar observations were made by Manzella,<sup>11</sup> Mittal<sup>12</sup> and Martini.<sup>13</sup>

'Atypical' mycobacteria have been the subject of more clinical awareness. The organisms are difficult to isolate and are resistant to the usual antituberculosis drugs. The lesions of fungal infection are usually 'space-occupying mass' induced erosions.<sup>1</sup> These include maduramycosis, coccidioidomycosis, histoplasmosis, blastomycosis and sporotrichosis. Lesions of sarcoidosis of the hands and feet are usually cyst-like.<sup>1</sup>

Neoplasms of the bone can mimic osteomyelitis. Such lesions are usually metastatic from tumours of the breast, lung or prostate. The lesions are permeative and do not have a spheroid appearance like actinomycosis.

Lesions of actinomycosis resemble multiple myeloma, are spheroid in nature, and multiple. The reactive bone margins of actinomycosis are not common in multiple myeloma.

## Conclusion

Actinomycosis is a rare disease encountered in orthopaedics. Calcaneal involvement has not been reported. The diagnosis can easily be overlooked for more common conditions with discharging sinuses, such as pyogenic osteomyelitis and tuberculosis. Bone biopsies taken at multiple sites and delivered to the laboratory as soon as possible in an anaerobic medium is advised.

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