



# Primary actinomycosis of the thigh – a rare soft tissue infection with review of literature

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## DECLARATIONS

### Competing interests

None declared

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### Ethical approval

Written informed consent was obtained from the patient or next of kin

### Guarantor

Rohan Khandelwal

### Contributorship

RK, PCM:

Operating surgeons and involved in manuscript preparation.

IJ: Pathologist;

reported the histo-pathology slides

SP, AS, SY:

Assisting surgeons and helped in manuscript preparation

PS, SS: Helped in review of literature

Primary actinomycosis is a rare soft tissue infection which requires a high degree of clinical suspicion to diagnose and it should be differentiated from other chronic conditions like tuberculosis.

## Case report

A 42-year-old lady presented to the surgical OPD with complaints of a swelling with multiple discharging sinuses over her right thigh for the past two years. The swelling was insidious in onset and was gradually increasing in size. There was no history of any trauma or insect bite at the site of the lesion. The patient had no past history suggestive of tuberculosis or diabetes mellitus. She was postmenopausal for the last four years and had no past history of intra-uterine contraceptive device insertion. She had been treated by multiple doctors in various centres, over the last 6 months and had taken extended courses of oral antimicrobial therapy.

On examination there was a 15 cm × 20 cm, irregularly shaped, indurated swelling in the medial aspect of the right thigh extending from the perineum to mid-thigh (Figure 1). There were multiple sinuses draining yellow coloured pus along with sulphur granules. There were no other lesions elsewhere on the body.

The pus from the discharging sinuses was sent for microscopy and a biopsy from the lesion was sent for histo-pathological examination. Gram staining revealed Gram positive bacilli, but no organisms could be cultured. Histopathological examination confirmed the diagnosis of actinomycosis (Figure 2).

A MRI scan was done and it revealed multiple sinuses in the medial aspect of the right thigh extending to the underlying muscles (Figure 3). There were no lesions in the underlying bone.

The patient was started on co-trimoxazole and cindamycin for a period of 1 month, after which the patient was taken up for surgical excision of the lesion followed by split skin grafting to cover the created defect (Figure 4). During excision, the whole lesion along with the involved muscle was removed. Multiple sinus tracks were encountered during surgery.

The patient had a wound infection in the postoperative period, which responded to parental antibiotics. The patient was discharged on oral co-trimoxazole and clindamycin for 15 days following which only co-trimoxazole was continued for 2 months.

The patient has been in regular follow-up and there has been no recurrence till after 1 year of surgery (Figure 5).

## Discussion

Actinomycosis is subacute to chronic, suppurative granulomatous disease that tends to produce draining sinus tracts. This usually manifests as cervico-facial, thoracic or abdominal actinomycosis. It is most commonly caused by *Actinomyces israelii* in humans and *Actinomyces viscosus*, *Actinomyces bovis*, *Actinomyces naeslundii* in animals. Actinomycosis is an anaerobic, gram positive bacterial infection which is seen in different parts of the body and exists as a saprophyte. It mostly occurs in patients belonging to low

and management of  
the patient  
  
Reviewer  
Jody Parker

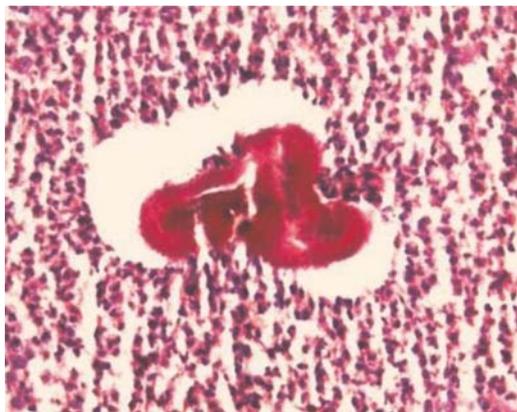
**Figure 1**  
Actinomycotic lesion over the thigh extending to the perineum. Multiple openings of sinuses can be seen



socioeconomic strata and having poor dental hygiene.<sup>1-3</sup>

*Actinomyces* produces disease when it is introduced into tissues by trauma, surgery, or infection. Actinomycosis commonly involves colon, mouth and vagina. Actinomycosis can occur at all ages of life with a peak in middle ages. Primary disease of the extremities is uncommon and has

**Figure 2**  
High power view (100X) showing an actinomycotic colony in a background of chronic inflammation



**Figure 3**  
MRI image showing multiple sinuses extending till the underlying muscles. The bone appears to be normal



an association with trauma and bites.<sup>1,4-6</sup> In our case, there was no history of past trauma or surgery and the actinomycosis was primary in nature.

Estimation of the exact incidence of actinomycosis is difficult, because there are probably many undiagnosed cases of actinomycosis. Actinomycosis has been called 'the most misdiagnosed disease' even by experienced clinicians, and listed as a 'rare disease' by the Office of Rare Diseases (ORD) of the National Institutes of Health (NIH).<sup>7</sup> The diagnosis is a challenge and

**Figure 4**  
Postoperative picture after resection of the lesion and split skin thickness graft in place



**Figure 5**  
Picture after one year of surgery, showing no signs of recurrence and a well healed scar



often delayed, with a protracted history of masses and sinuses extending into the gluteal and genital region.<sup>8</sup>

The infection begins as an inflammatory soft tissue mass, which can enlarge into an abscess like swelling, with penetration of the overlying skin which mimics malignancy. Cutaneous actinomycosis manifesting with nodular lesions that tend to form fistulae needs to be differentiated clinically from other chronic inflammatory skin diseases, such as cutaneous tuberculosis, sporotrichosis, and nocardiosis.<sup>9</sup>

Diagnosis requires high suspicion index. Diagnosis is based on identification of sulphur granules (usually 1–6 mm in diameter) in pus and histological preparations, and preferably also by a positive culture.<sup>8</sup>

Histology is diagnostic, characteristically showing colonies (sulphur granules) formed by tangled mass of filaments surrounded by radiating, sometimes terminally clubbed, organisms.<sup>9</sup> The bacteria are filamentous, gram-positive, anaerobic-to-microaerophilic and are not acid fast.<sup>8</sup>

There is no standardized treatment of actinomycosis mentioned in the literature. It involves a combination of surgery and antibiotics, and is poorly standardized.<sup>5</sup> Crystalline penicillin is widely used in the management of such lesions, and other drugs such as ampicillin/amoxicillin, erythromycin, sulphonamides, streptomycin, chloramphenicol, other tetracyclines and rifampicin have also been used.<sup>10</sup>

Surgery is indicated for small lesions or those which do not respond to medical treatment. There is a role of prolonged course of antibiotics after surgery.

#### References

- 1 Roy Debabrata, Roy Pankaj Gupta, Misra PK. An interesting case of primary cutaneous actinomycosis. *Dermatology Online Journal* 2003;9:17
- 2 Ermis I, Topalan M, Aydin A, Erer M. Actinomycosis of the frontal and parotid regions. *Ann Plast Surg* 2001;1:55–58
- 3 Sardana K, Mendiratta V, Sharma RC. A suspected case of primary cutaneous actinomycosis on the buttock. *J Dermatol* 2001;28:276–8
- 4 Wee SH, Chang SN, Shim JY, Chun SL, Park WH. A case of primary cutaneous actinomycosis. *J Dermatol* 2000; 27:651–4
- 5 Metgud SC, Sumati H, Sheetal P. Cutaneous actinomycosis: A rare case. *Indian J Med Microbiol* 2007;25:413–5
- 6 Daie Ghazvini R, Zaini F, Zibafar E, Omid K. First case report of primary actinomycosis of the breast due to *Actinomyces israelii* from Iran. *Acta Medica Iranica* 2003;41:111
- 7 Schall KP. Actinomycosis, Actinobacillosis and related diseases. In: William J, Hausler MS Jr eds. *Topley and Wilson's Microbiology and Microbial Infections*, vol. 3. Bacterial Infections, 9th ed. Arnold: Great Britain; 1998:777–98
- 8 Coremans G, Margaritis V, Van Poppel HP, et al. Actinomycosis, a rare and unsuspected cause of anal fistulous abscess: report of three cases and review of the literature. *Dis Colon Rectum* 2005;48:575–81
- 9 Reiner SL, Harrelson JM, Miller SE, Hill GB, Gallis HA. Primary Actinomycosis of an extremity: A case report and review. *Rev Infect Dis* 1987;9:581–9
- 10 Manas Chatterjee, Gluteal primary cutaneous actinomycosis, *Indian Journal of Dermatology* 2005;50:152–4

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