

Photoclinic



Figure 1. Multiple nodular lesion and sinus tracts on the planter surface of the right foot.

A 61-year-old woman presented to the Infection Ward of Imam Khomeini Hospital with right foot edema, multiple nodular lesions, and sinus tracts (Figure 1). The lesions were mainly distributed on the plantar surface with some extension onto the dorsal surface. The initial lesion was a single nodule, followed by the appearance of successive nodules in close proximity. This problem began five years prior and progressed gradually. At presentation, she was a febrile and her right foot was firm with nontender swelling. On examination, the right foot showed swelling and induration. The surface of her foot showed multiple nodules with draining sinuses and crusting. The skin was hypo- and

hyperpigmented with signs of both old healed and active sinuses. A few granules were present in the active sinuses. X-ray of the foot was normal. The patient, a farmer, was from a northern Iranian village. Her foot had been exposed to contaminated soil.

Abdulreza Suodbakhsh MD¹, Alireza Soleimani MD*¹, Hamid Emadi MD¹

Authors' affiliation: ¹Department of Infectious Diseases, Imam Khomeini Hospital, Tehran University of Medical Sciences, Tehran, Iran.

•Corresponding author and reprints: Alireza Soleimani MD, Department of Infectious Diseases, Imam Khomeini Hospital, Tehran University of Medical Sciences, Tehran, Iran. Tel: +98-912-245-4275, Fax: +98-216-658-1598, E-mail: ali.doctor@yahoo.com

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**What is your diagnosis?
See the next page for diagnosis.**

Upon admission, our clinical diagnosis was mycetoma. Mycetoma is a disease of localized, indolent, deforming swollen lesions and sinuses involving cutaneous and subcutaneous tissues, fascia, and bone. The etiologic agent can be a variety of bacteria (Actinomycotic mycetoma or Actinomycetoma) or fungi (Eumycotic mycetoma or Eumycetoma).¹ Because drug therapy varies for actinomycetoma and eumycetoma, the clinician must differentiate between actinomycetes or fungi. This diagnosis was confirmed by gram staining and culture of the secretion, which revealed a higher bacteria consistent with actinomycetes. A smear for acid fast bacilli was negative. Fungal culture did not yield any growth. Biopsy showed the presence of micro-abscesses in the subcutaneous tissue composed of neutrophils, eosinophils, plasma cells and, macrophages. The center of these tissue contained colonies of actinomycetes.

Actinomycetoma is caused by members of the order Actinomycetales, most commonly *Nocardia brasiliensis*, *Actinomadura madurae*, *Streptomyces somaliensis*, and *Actinomadura pelletieri*. A few cases have been reported caused by *Actinomadura latina*, *Nocardia asteroides*, *Nocardia otitidiscaviarum*, *Nocardia transvalensis*, and *Nocardiaopsis dassonvillei*.² Actinomycetoma granules are white or pale yellow, except those caused by *Actinomadura pelletieri*, which are red to pink.¹ The prevalence of mycetomas and infective agents in Iran between 1972 and 2005 have been discussed in a review article. Seventy-six cases of mycetomas have been reported from various geographical locations in Iran during the past 33 years. Analysis of the records revealed that 84.5% were actinomycetoma with only 15.5% diagnosed as eumycetoma. The disease mainly presents in the feet with a male to female ratio of 2:1. Mycetomas are abundant among farmers in rural areas of Iran. The most common agents of mycetomas have been *Nocardia asteroides*, *Actinomadura madurae* (actinomycetoma), and *Allesheria boydii* (eumycetoma). The peak age of onset was between 31 and 51 years.³

The etiologic agents of mycetomas are a group of saprophytic fungi and actinomycetes that live in soil. The disease results from the traumatic implantation of soil organisms into the tissues, most often affecting the lower extremities, typically a single foot.³

This disease may also result from decreased use of protective clothing, chiefly shoes in the warmer, poorer endemic regions. In most cases, this disease begins as a single, small, painless subcutaneous nodule that slowly increases in size, becomes fixed to the underlying tissue and ultimately develops sinus tracts beneath the lesion. These tracts open to the surface and drain purulent material with grains. Grains are several millimeters in diameter and may be seen by close inspection of a gauze bandage that is placed over the sinus tract. Progression to draining sinus tracts can take weeks, months, and even years, occurring more rapidly in actinomycetoma.¹ The disease can affect the skin, subcutaneous tissue and eventually contiguous bone, spreading along fascial planes. Muscle, tendons, and nerves are generally spared direct infection, but extensive local damage may lead to muscle wasting, bone destruction, and limb deformities. Hematogenous spread has not been documented and thus no signs or symptoms of systemic illness are usually seen in mycetoma, unless a secondary bacterial infection occurs. When left untreated, the disease continues to progress, and bacterial superinfection can lead to increased morbidity from local abscess formation, cellulitis, bacterial osteomyelitis and rarely, septic death.

Differential diagnosis includes botryomycosis, chronic bacterial osteomyelitis, and chromoblastomycosis.¹

Diagnosis of mycetoma can be made by the classic triad of painless soft tissue swelling, draining sinus tracts and extrusion of granules. Diagnosis of the causative organism can either be made by microscopic observation and culture of a granule recovered from aspirated material or biopsy specimens used to diagnose the specific cause of mycetoma. Deep biopsy with histopathology and culture is usually not necessary.¹

Treatment regimens are currently based on expert opinion as no randomized, controlled trials have been performed. Duration of therapy is also not defined, with most patients receiving 3 to 24 months of therapy to affect an adequate response.¹ Prognostically, actinomycetoma can be cured with surgical debridement and appropriate antibiotic therapy while eumycetoma is only partially responsive to antifungal agents, has a high rate of recurrence and may require amputation.

The most commonly described regimens for actinomycetoma include streptomycin plus either trimethoprim-sulfamethoxazole (TMP-SMX) or dapsone.¹ In this regimen, streptomycin (14 mg/kg/day) is given intramuscularly for the first month (and sometimes three times weekly thereafter for several months) in addition to a long course of TMP-SMX, usually one double-strength tablet (160 mg trimethoprim and 800 mg sulfamethoxazole) twice daily or dapsone (1.5 mg/kg/day twice daily). Alternate regimens include TMP-SMX plus dapsone and amikacin plus TMP-SMX.⁴ Cycled dosing of amikacin (15 mg/kg/day divided into two daily doses for three weeks) in addition to TMP-SMX for five weeks has also been administered.⁵ Most patients improve with only one to two cycles of this therapy. Response to TMP-SMX alone has also been reported.⁶ Other regimens that have been used include streptomycin with either sulfadoxine-pyrimethamine or rifampin; a combination of penicillin, gentamicin, and TMP-SMX followed by amoxicillin and TMP-SMX,⁷ and regimens that include amoxicillin-clavulanate,⁸ fusidic acid, clindamycin, or imipenem/cilastatin.

Our patient was administered streptomycin plus TMP-SMX. In this regimen, streptomycin (14 mg/day) was given intramuscularly for the first month, in addition to a long course of TMP-SMX (one double strength tablet twice daily).

Disease prevention is best accomplished by wearing shoes and clothing to protect against splinters and thorn picks.¹

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