

## Case Report

# Recurrent madura foot without draining sinuses: a case report

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### ABSTRACT

Madura foot or mycetoma is a chronic granulomatous disease characterized by localized infection of subcutaneous tissues by actinomycetes or fungi. The recurrence rate for the disease if treated inadequately is very high. Recurrence presents with swelling and multiple discharging sinuses. This is an unusual presentation of the disease without discharging sinuses which is probable the first report of this kind in the literature. A 34 year old, male, presented with the painless, progressive swellings over right foot since 4 years. No sinuses or discharge could be found on skin surface. The postoperative recurrence rate is very high, and this can be local or distant at the regional lymph nodes. This could be due to the disease biology and behavior or inadequate surgical excision. Usually it presents with multiple sinus tracts, and granule. We reported a case with classical absence of sinus tracts in recurrent actinomycosis.

**Keywords:** Actinomycosis, Recurrent, Madura foot, Draining sinuses

### INTRODUCTION

Madura foot is a chronic infection of the skin and underlying tissues caused by both bacteria (actinomycetomas) and fungi (eumycetomas). They are characterized by indolent tumefaction of the affected area, multiple sinus tracts, and granules that contain the agent.<sup>1</sup> It is endemic in the tropics and subtropics. It is a deep mycosis caused by exogenous fungus or actinomycotic species. These infections lead to progressive inflammation of the skin, subcutaneous tissue, muscles and bones. The organism enters through local trauma in the foot, hand or eyes from saprophytic soil. After entry to the body they form subcutaneous nodules containing suppurative granulomas, multiple cavities and sinus tracts. The sinus tracts discharge exudates with fine grains. These grains are colonies of causal organism.<sup>2</sup> The recurrent disease presents with discharge and sinuses. Here is an interesting

presentation of recurrent actinomycosis with classical absence of sinus tracts.

### Review of literature

The disease was first described in the Indian town of Madura. Mycetoma occurs most often in people, who work in rural areas, usually in farmers, hunter-gatherer populations, and field labourers where they are exposed to acacia trees or cactus thorns that contain the etiologic agents that normally live as saprophytes. The disease can also be found in people who work in the city in various occupations, in victims of road accidents who have incurred a traumatic inoculation of the agent, and in travelers to tropical endemic areas. Subtropical regions and the African continent have the highest prevalence. Eumycetoma prevails in the mycetoma belt that stretches between the latitudes of 15° south and 30° north.<sup>3</sup> Due to

the existing socio-economic condition and low living standard of the people of this area the disease is often neglected in the initial stage. It may occur in all age groups.

The diagnosis is made at an advanced stage and good clinical response with proper pharmacological therapy alone has been reported.<sup>4</sup> Surgical debridement long with antifungal and antibiotic has been proved effective in many cases. Amputation of limbs followed by antifungal and antibiotic therapy and reconstruction have been done for a number of cases.<sup>5</sup>

Although it is not an opportunistic infectious disease, it has been associated with the use of corticosteroids, leukemia and children with congenital immunodeficiencies and HIV infection. The 3 major forms of actinomycosis are cervicofacial (65%), thoracic (15%) and abdominal/pelvic (20%)<sup>6</sup>. A severe form of periodontitis may be associated with actinomyces. It has propensity to infect the heart valve and thus cause the insidious presentation of endocarditis with fever in less than half of the cases.<sup>7</sup> Hand and foot are the rare sites of involvement and it is reported in other sites also, in various studies.

## CASE REPORT

A 34 year old, male patient, presented to our hospital, Acharya Vinoba Bhave Rural Hospital, with the complaints of multiple swellings over the right foot since 4 years. It was painless but causing discomfort while walking because of its location. The progression was slow. No sinuses or discharge could be found on skin surface. General condition of the patient was fair. On Local examination three swellings were present over the right foot, size ranging from 6-8 cms, nontender, and firm in consistency, with irregular surface. No sinuses or discharge was present. All biochemical tests were within normal limits. No bony abnormality but soft tissue shadows were present on X ray foot. Intra-operative, multinodular, friable growth involving whole of the plantar surface and extending to both the malleoli was resected. However classical sinuses were not seen.



**Figure 1: Preoperative photograph with classical absence of discharging sinus tracts and scar of previous surgery.**



**Figure 2: Intra-operative photograph showing multiple, nodular swellings.**

## DISCUSSION

Actinomycosis, which was first described by Israel in 1878,<sup>8</sup> is a rare, chronic, spreading, suppurative, granulomatous and fibrosing infection characterized by the formation of multiple abscesses, draining sinuses and the release of characteristic “sulfur granules”. It is found worldwide and occurs at any age, but is rare at ages younger than ten. The peak incidence is between 15 and 30 years, and males are more frequently infected than females. Madura foot was first recognized in 1842 by Gill in Madura district of Tamilnadu in India.<sup>3</sup> Later Bidie and Carter gave a full description of the disease.<sup>9</sup> Mycetomas are frequent in the tropical zones of America (Mexico and Venezuela), Africa (Senegal, Mauritania and Sudan) and in India, but can also be observed in other areas. These Fungi in rainy areas are found as saprophytes in the soil, are usually introduced through skin wounds in those who walk bare footed generally farmers and nomads who are susceptible for exposure to penetrating wounds.

Infection begins in the skin and subcutaneous tissue. Pathologically lesions appear first as local papular or nodular swelling which tends to grow and rupture, forming communicating sinus tracts through which mucous containing the characteristic colored grains is discharged. Some sinuses heal over the time and new are formed with the areas of scarred tissues leaving behind; gradually the affected part enlarges and becomes disfigured. The infection gains entry into the bone and replace the spongiosa. The fever is indicative of secondary bacterial infection in the pre-existing lesion. The infection does not spread hematogenously. Some cases are known where particularly *Pseudallescheria boydii* and *Nocardia asteroides* in immunocompromised patients (leukemia, HIV, use of corticosteroids and immunosuppressivedrugs) have disseminated hematogenously to the brain, myocardium and the thyroid gland.<sup>10</sup>

Actinomyces are gram-positive, non-acid fast, anaerobic or microaerophilic filamentous branched bacteria which are very difficult to grow in culture, with <30% of

cultures being positive. In man, the pathogenic Actinomyces most frequently isolated is *A. Israelii*; less commonly, infection is caused by *A. Propionica*, *A. Naeslundii*, *A. Viscosus* and *A. Odontolyticus*. These bacteria are all normal commensals of the human oral cavity.<sup>11</sup>

The treatment of mycetoma depends mainly on its etiological agent and the extent of the Disease. Until recently, the only available treatment for mycetoma was amputation of the affected part or multiple mutilating surgical excisions. No case of self cure has ever been reported in the medical literature. However, spontaneous lesion regression was observed in some patients. Combined medical and surgical treatment is the gold standard in eumycetoma.<sup>12</sup> In general, actinomycetoma is amenable to medical treatment with antibiotics and other chemotherapeutic agents. Combined drug therapy is always preferred to a single drug to avoid drug resistance and for disease eradication. In actinomycetoma, combined medical and surgical treatments are beneficial. This regime facilitates surgery, accelerates healing and reduces the chance of relapse; however, a good number of patients respond to medical treatment alone.<sup>13</sup>

Medical treatment is effective in all stages of actinomycetoma, even in patients with advanced disease and with a great deal of bone damage. Cure is possible, although a prolonged period of treatment is needed. Recurrence is more common after an incomplete or irregular course of medical treatment. Medical treatment for both types of mycetoma must continue until the patient is clinically, radiologically, ultrasonically and cytologically cured. Cure is considered when the skin becomes normal, the mass disappears, the sinuses heal and the organisms are eliminated from the tissue. Clinical improvement is judged by reduction in the size of the mass and healing of most of the sinuses.<sup>12</sup>

Surgery is indicated in mycetoma for localized lesions, resistance to medical treatment or for better response to medical treatment in patients with massive disease. The surgical options range from wide local and debulking excisions to amputations. Amputation is indicated in advanced mycetoma not responding to medical treatment with severe secondary bacterial infection, and it can be a life-saving procedure. The amputation rate ranges from 10-25% in most series.<sup>12,13</sup> The postoperative recurrence rate varies from 25 to 50%, and this can be local or distant at the regional lymph nodes. This could be due to the disease biology and behavior, inadequate surgical excision due to the use of local anesthesia and lack of surgical experience and drug compliance due to financial reasons and/or lack of health education.<sup>13</sup>

The current most acceptable treatment options for actinomycetoma are cycles of the combination of amikacin sulfate 15 mg/kg twice daily for 3 weeks and cotrimoxazole 1.5 mg/kg twice daily for 5 weeks until cure. For eumycetoma, ketoconazole 800 mg/day is the

recommended drug; however, for patients who show poor response and/or developed severe side effects, itraconazole 400 mg/day is a good option. The mean duration is 9 months. Patients should be closely followed up clinically and biochemically for the drug's side effects.<sup>12,13</sup>

The morbidity caused by mycetoma is extensive, and in areas where mycetoma is endemic, local healthcare facilities and health education are usually insufficient and inadequate. Mycetoma is a hard-to-treat disease in many tropical and subtropical regions. The mycetoma clinical and socio-economic impacts have essentially remained unchanged over the last decades. However, these drugs proved to be expensive to patients and health authorities, remained limited to certain patients by the poor availability, and have many serious side effects.

## CONCLUSION

We reported a case of recurrent actinomycosis without classical discharging sinus tracts. This may either mislead or delay the diagnosis. Careful history and examinations to rule out other soft tissue tumors are sought for.

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