

A Case of a 62-Year Old Male with Actinomycetoma Successfully Treated using Combined Medical and Surgical Therapy

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Abstract

Introduction: Actinomycetoma is a neglected infectious disease that presents with a triad of subcutaneous mass, with sinus formation and seropurulent discharge containing grains. This causes severe functional disability to patients if not treated early and properly.

Case Report: We report a case of a 62-year-old adult Filipino farmer diagnosed with actinomycetoma of the left foot. For 8 years, the patient medicated with several antibiotic therapies affording minimal improvement of symptoms. Hence, surgical intervention with combination antimicrobial therapy was done which provided resolution of symptoms, improving the quality of life of the patient.

Conclusion: Treatment of actinomycetoma must be individualized among patients. Although oral antibiotics became the standard of treatment, combining surgical treatment with oral medications may be considered to ensure effective management of the disease.

Key Words: actinomycetoma, cotrimoxazole, dapsone, surgery, case report

INTRODUCTION

Actinomycetoma is an infectious disease caused by the bacteria, *Nocardia* sp., *Streptomyces* sp., or *Actinomyces* sp. It commonly affects agricultural workers in developing countries in tropical and subtropical zones including the Philippines. It is characterized by chronic inflammation with discharge and sinus formation. Actinomycetoma continues to pose huge public health threat in many countries since its course can be quite devastating ^[1].

There is no proposed algorithm in the management and treatment of Actinomycetoma. Currently, the disease is treated with prolonged course of antibiotics. However, in some instances, medical cure even with prolonged combination of antibiotics is difficult to achieve. Extension of the disease may lead to chronic progressive lesions and sometimes gruesome deformity of the limbs. This causes significant life-long disabilities and pain. It may also provide physical and socio-economic burden on the patient and the family members ^[2].

CASE REPORT

A 62-year-old Filipino male who worked as a farmer in Pangasinan, Philippines presented with an 8-year history of swelling on the dorsum of the left foot associated with multiple nodules, seromucoid discharge and whitish grains after a penetrating injury by a thorn from a tree trunk. Several consultations were done, and he was given numerous unrecalled oral medications, which did not provide resolution of the lesions. Excision biopsy revealed Mycetoma (Maduromycosis) of the left foot leading to referral to a specialty center where he was treated as a case of Actinomycetoma. The patient was immediately given Trimethoprim-Sulfamethoxazole (TMP-SMX) 800mg+160mg tablet three times a day continuously for 4 years with good compliance to medications. However, the patient opted to use a generic brand due to financial difficulty, which caused the reactivation of the disease. He was advised to return to TMP-SMX with free Dapsone 50mg/tablet to be taken once daily for one year.

Cutaneous examination revealed multiple, well-defined, firm, fixed, hyperpigmented nodules, with thick central scaling and sinus tracts on the dorsal aspect of

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the left foot. There was no discharge or grains noted (Figure 1). Baseline laboratory tests such as complete blood count, urinalysis, blood chemistry and chest x-ray were all within normal limits. An x-ray of the left foot was done which revealed cortical irregularities and increased opacification with associated interspersed cystic lucencies of the 4th metatarsal bone, as well as cortical thickening of the 3rd and 5th metatarsal bones, suggestive of osteomyelitis (Figure 2).



Figure 1. Cutaneous examination of the left foot



Figure 2. AP and Oblique views of the left foot

Due to the prolonged intake of oral medications, a wide excision biopsy was done. An irregularly shaped tissue with superficial white plaques measuring 8.0 x 5.0 x 0.4 cm was obtained (Figure 3). Intraoperatively, sinus tracts with whitish to yellowish discharge were observed. Hematoxylin and eosin (H&E) staining of the tissue revealed a normal epidermis with pigmented basal layer, areas with pseudoepitheliomatous hyperplasia with densely packed collagen and infiltrates of histiocytes suggestive of a scar tissue. Gram stain and culture of the discharge revealed no growth after 5 days of incubation.



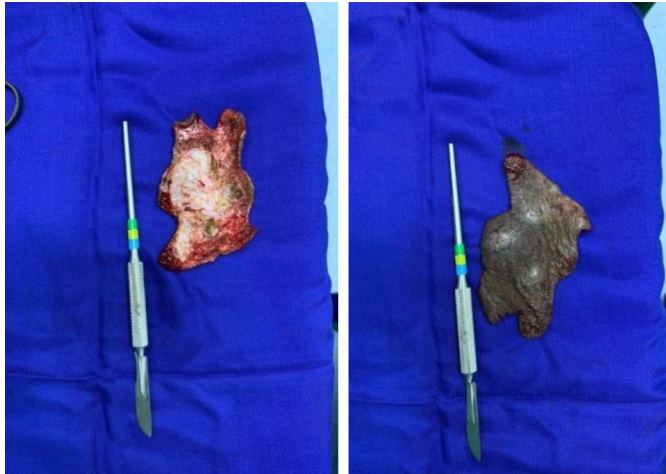


Figure 3. Intraoperative findings during wide excision biopsy

After surgery, TMP-SMX was continued for another 3 months. At 3 months of follow up, there were no new lesions observed. No new growths of nodules with sinuses were noted (Figure 4). The patient had ease of pain on ambulation with no difficulty in executing normal activities of daily living.



Figure 4. Clinical improvement after surgical and medical management

DISCUSSION

Actinomycetoma has been included in the top 17 list of neglected tropical disease by the World Health Organization in 2013 [3]. As of 2015, there were only 97 reported cases seen in the Philippines. Although the incidence of actinomycetoma is rare in the country, this

disease causes disfiguring lesions in affected individuals, limiting their mobility and affecting their quality of life [4].

The triad of subcutaneous mass, multiple sinuses and discharge with whitish grains were suggestive of actinomycetoma [5]. Risk factors such as traumatic injury, walking barefoot on land and the low socioeconomic status of the patient increases the likelihood of the disease.

Diagnostic procedures such as direct examination of the grains from the sinuses, special stains and biopsy may be done to confirm the diagnosis. However, H&E staining of the mass on the left foot shows fibrosis and scar tissue even though the clinical picture was suggestive of actinomycetoma. Gram stain and culture of the discharge from the sinuses were also negative. According to Tahiri, et al, in long standing disease of actinomycetoma, inadequate amount of grains and granules may be apparent if the patient had multiple prior treatments such as in the case of our patient wherein he was taking antibiotics for 4 years [6]. This is supported by the case report of Koley, et al which suggested that late presentation of the disease may show mainly fibrosis and scarring on histopathology. The extent of the fibrosis can make the discharge from the sinuses inapparent [7]. These factors may reveal non-specific findings on histopathology. Therefore, in such cases, it is important for the histopathological report to be correlated clinically.

A review of related literature states that there is no proposed algorithm for its treatment, and therapy is individualized with antibiotic treatment as first line [8]. The latest systematic review done by Scolding, et al. shows that TMP-SMX is the first line drug with minimum expected duration of treatment for 12 months [9]. The disappearance of the swelling, complete healing and closure of sinuses, absence of evidence of mycetoma on histology and/or culture, and absence of disabilities secondary to pain or deformity are the criteria for clinical cure [9].

Our patient has been taking these medications for 4 years but has not reached the clinical criteria for cure. Thus, wide excision biopsy was done to debulk the

lesion, debride all damaged tissues and remove all possible sinuses. Studies have shown that antibiotics combined with surgical procedures have the best therapeutic response [10]. Surgical procedures such as debridement, excision and amputation are useful in extensive infections and those with failed medical therapy. Surgery is done to reduce the bacterial load, provide a better response to therapy and prevent recurrence of the disease. The systematic review conducted by Salim, et al recommended antimicrobials in conjunction with surgery in successfully managing actinomycetoma patients [8].

CONCLUSION

Actinomycetoma is a unique forsaken disease that has a profound and negative impact on the physical, emotional and socio-economic aspects of the lives of the patients, their families and their communities. Most often, it is seen amongst the poorest of people who have less access to healthcare. In a third world country like the Philippines where the chronic and progressive nature of the disease requires a prolonged and consistent intake of medications, sustainability of treatment is a challenge that we need to overcome in order to ensure treatment success. Thus, treatment must be individualized among patients. Although oral antibiotics became the standard of treatment for actinomycetoma, combining surgical treatment with oral medications may be considered to ensure effective management of the disease.

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