Actinomycotic mycetoma of the scalp managed in a resource-limited setting

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Actinomycotic mycetoma or actinomycetoma is a type of mycetoma caused by *Nocardia* and *Actinomyces*. It usually affects the trauma-prone areas or extremities of the body. It is associated with characteristic discharging granules and sinuses. Gram staining of discharged granules shows thin Gram-positive filaments. Histopathology of granules shows suppurative granulomas composed of neutrophils surrounding characteristic grains. Several antibiotics are effective, including co-trimoxazole, dapsone, streptomycin, trimethoprim (TMP), rifampicin, and amoxicillin-clavulanic acid. However, co-trimoxazole remains the gold-standard therapy. Actinomycetoma of the scalp has been rarely reported. Here, we report the case of an Asian male in his 50s presenting with swelling over the scalp. It had developed seven years ago after a road traffic accident and remained elusive to treatment. There were no discharging sinuses or granules characteristic of actinomycetoma. The Gram stain and Modified Ziehl-Neelsen stain from superficial swabs were negative for pathogens. Bacterial and fungal cultures of the biopsy sample were inconclusive. However, histopathology showed epidermis having focal acanthosis overlying granulation tissue with proliferating capillaries, edema, and infiltration by lymphocytes, plasma cells, neutrophils, and eosinophils along with scattered foreign body giant cells. Filamentous bacterial colonies with surrounding neutrophils were present. These features were suggestive of actinomycetoma. The patient was treated with oral doxycycline and co-trimoxazole and had a complete regression of swelling after three months of follow-up. This case highlights an unusual morphology and location of actinomycetoma, which should be considered when encountering subcutaneous swelling.

Keywords: mycetoma, nocardia, trimethoprim, sulfamethoxazole, drug combination, actinomyces, neutrophils

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INTRODUCTION

Actinomycetoma is a type of mycetoma with characteristic granular discharge and sinuses involving mostly the extremities [1–4]. It rarely involves the head and neck region [4–7]. Gram staining of discharged granules shows thin gram-positive filaments, which are partially acid-fast bacilli [2]. Histopathology features include the presence of suppurative granulomas composed of neutrophils surrounding characteristic grains in the subcutaneous tissue. Grains appear as broad, pink-stained hyphae surrounded by a sharp basophilic strand [1–3]. Cultivation of *Actinomyces*

requires incubation for a prolonged duration (ten days) on special media like Lowenstein–Jensen (LJ) media, thioglycollate broth, Columbia agar, or brain heart infusion agar [1,3]. Various treatment regimens have been developed, but co-trimoxazole is the mainstay of treatment [1,6–9].

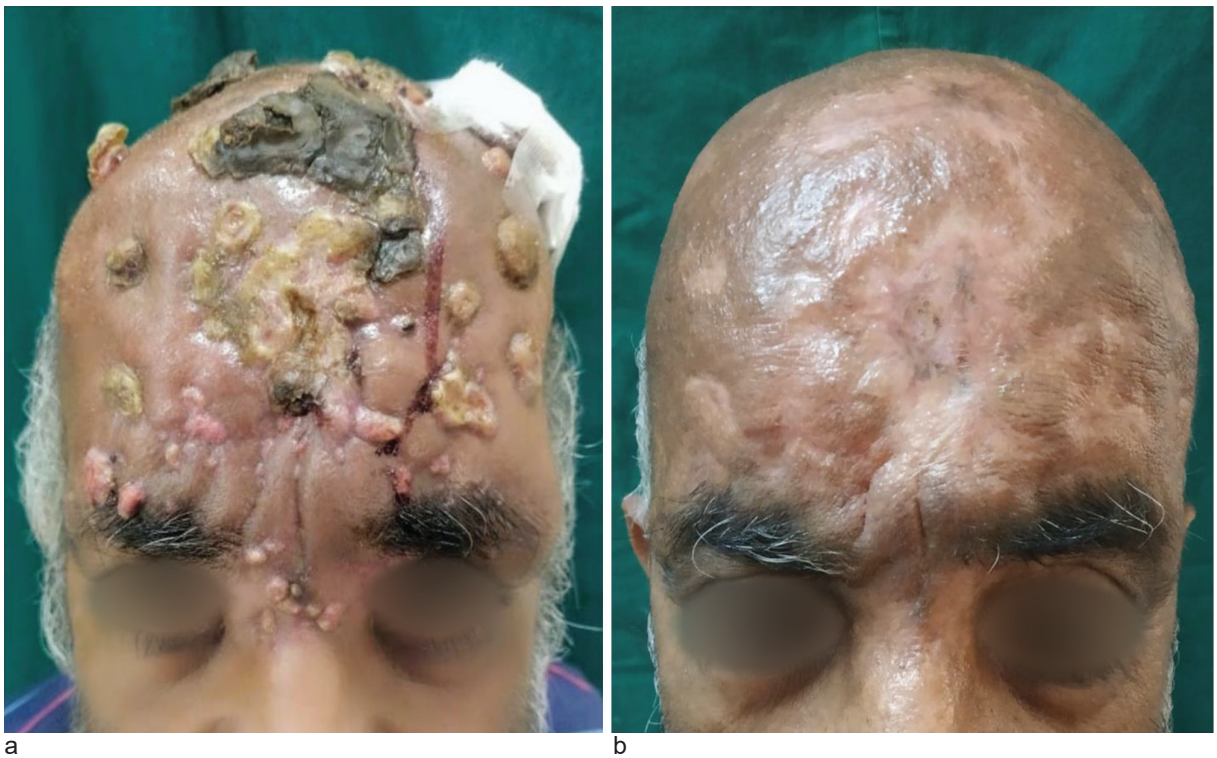
We report a case of actinomycetoma involving the head without the typical discharging sinuses and grains. The patient was elusive to treatment due to his tribal location. Histopathological features confirmed the presence of actinomycetoma, and he was treated accordingly.

CASE PRESENTATION

An Asian male in his 50s developed a localized swelling on the scalp, progressively increasing over seven years after a road traffic accident leading to injuries to the scalp. He did not receive treatment for his condition due to his tribal location and financial limitations. However, he used native herbal pastes of unknown composition for many years.

At the time of presentation, the swelling extended from the root of the nose and inner canthus of eyes anteriorly till 10 cm from the vertex posteriorly and temple areas laterally with a firm consistency (Figure 1a). It had multiple scattered vegetative growths exuding foul-smelling non-granular yellowish discharge with overlying crusts. Discharging granules and sinuses were absent after saline compress for 24 hours. Some areas showed hemorrhagic crusting. The insinuation sign was negative. Systemic complaints like fever, headache, and respiratory symptoms were absent. General physical examination was normal. Complete blood counts (CBC) and liver and renal function tests were normal. HIV serology was negative; no signs or symptoms of immunosuppression were seen.

Gram staining and modified Zeihl-Neelson (ZN) staining from superficial wound swabs were negative for pathogens. Incisional biopsy was done for histopathology and fungal culture. Fungal and bacterial cultures on LJ media showed no growth after seven days of inoculation. Histopathology revealed epidermis showing focal acanthosis overlying granulation tissue with proliferating capillaries, edema, and infiltration by lymphocytes, plasma cells, neutrophils, and eosinophils along



a b

Figure 1. Clinical image on presentation (a) and on three-month follow-up (b).

with scattered foreign body giant cells. Filamentous bacterial colonies with surrounding neutrophils were present (Figure 2). The modified ZN stain was negative. Gram-positive and basophilic fine filamentous Periodic Acid Schiff (PAS) positive colonies were seen (Figure 3a). Gommori's Methenamine Silver (GMS) stain showed slender filamentous threads (Figure 3b). X-ray of the skull revealed multiple lytic lesions in the skull with cortical bone involvement. The histopathological findings prompted the diagnosis of actinomycotic mycetoma. The patient was treated with doxycycline 100 mg twice daily and co-trimoxazole 960 mg twice daily and had a complete regression of the swelling after three months of treatment (Figure 1b).

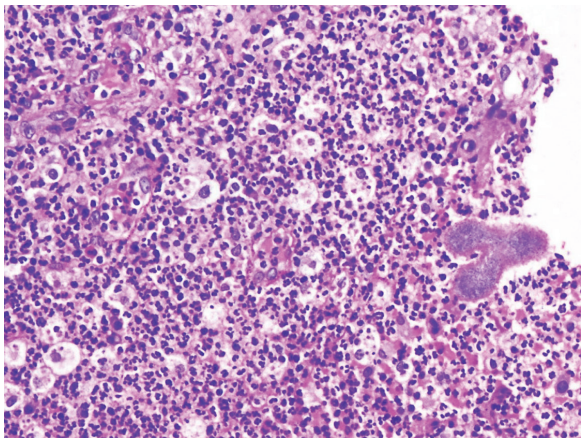


Figure 2. Photomicrograph shows purple colonies surrounded by granulation tissue with proliferating capillaries and dense infiltrate of neutrophils and macrophages (H&E, ×200).

DISCUSSION

Mycetoma can be classified as eumycetoma (due to true fungus), actinomycetoma (due to *Actinomyces*), and botryomycosis (mainly due to *Staphylococcus aureus*) [1–3,5,10]. Actinomycotic mycetoma or actinomycetoma is caused by *Actinomyces* (mainly *Actinomadura madurae* and *Actinomadura pelletieri*), *Streptomyces somaliensis*, and Nocardia (mainly *Nocardia brasiliensis* and *Nocardia asteroids)* [1–3]. The pathognomonic feature of a mycetoma is a painless, firm subcutaneous mass with multiple sinus formations, discharging purulent or seropurulent exudate that contains grains; this is similar across all its subtypes [1,3]. Over 75% of mycetoma cases are seen in the lower extremity, most commonly in the foot (70%) followed by the hand, rarely affecting the head and neck region [2,4–7,9].

In the case of actinomycetoma, Gram staining of discharged granules shows thin Gram-positive filaments, which are partially acid-fast bacilli. Histopathology shows the presence of suppurative granulomas composed of neutrophils surrounding characteristic grains, which are present in the subcutaneous tissue. Grains are microscopy visible

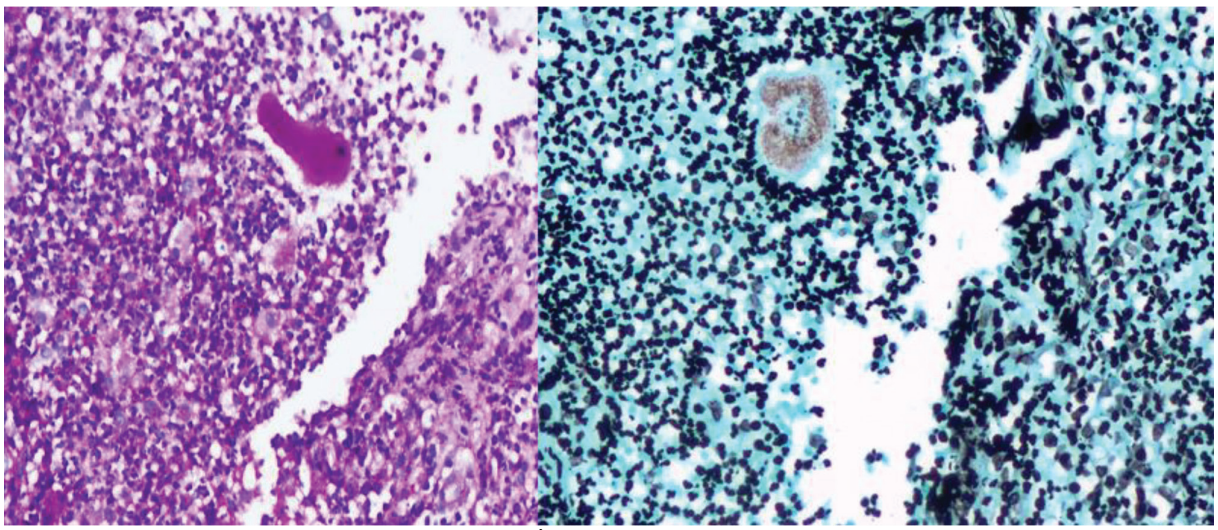


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Figure 3. Photomicrograph shows colonies that are positive for the PAS (a) and methenamine silver (b) stains, demonstrating fine filamentous colonies (×200).

as broad, pink-stained hyphae surrounded by a sharp basophilic strand. The neutrophilic infiltrate is further surrounded by palisading histiocytes, beyond which there is a mixed inflammatory infiltrate comprised of lymphocytes, plasma cells, eosinophils, and macrophages [1,2,4]. Cultivation of actinomycetes requires special media like LJ media, thioglycollate broth, Columbia agar, or brain heart infusion agar for a longer duration than other bacterial cultures (10 days) [1,2].

Common differentials for actinomycetoma include eumycetoma and botryomycosis [1]. Eumycetoma is due to fungal etiology, with a similar clinical presentation. They have thicker Gram-negative hyphae, stain positive with PAS and GMS, and produce fungal colonies on culture media after 4-6 weeks of inoculation [1]. Botryomycosis is a bacterial infection most commonly due to *S. aureus*, occurring most often in immunocompromised patients. It manifests mainly as subcutaneous nodules, verrucous lesions, or nonhealing ulcers associated with draining sinuses and fistulae draining purulent yellowish "grains" and exudate similar to actinomycosis. Microscopy shows Gram-positive cocci ≤1 micron in diameter surrounding the granule singly, in pairs, or in small clumps [1,10].

In our patient, the characteristic discharging sinuses and granules associated with the swelling were absent. Mycetoma usually involves the extremities. However, on a literature search, we came across the unusual presentation of actinomycetoma involving the head and neck region in the same region as ours [4,6,7,9]. Gram staining and modified ZN staining for AFB were performed from the superficial wound swab but did not reveal any pathogenic organism. One-week fungal and bacterial cultures were not sufficient to grow the pathogen. This limits our observation to a certain extent. However, histopathological features were suggestive of actinomycotic mycetoma.

Actinomycetoma is usually responsive to antibiotic treatment. Many antibiotics, like co-trimoxazole, dapsone, streptomycin, trimethoprim (TMP), rifampicin, and amoxicillin-clavulanic acid, have been used and found to be effective [3,4,8,9]. Common consensus is that co-trimoxazole should be administered as a gold standard therapy in all actinomycetoma patients. Combination antibiotic therapy is preferable to monotherapy to avoid the development of drug resistance and to eradicate residual infection [8].

The Welsh regimen includes cyclical dosing of amikacin 15 mg/kg/day, in two divided doses in cycles of 21 days for 1–3 cycles with intervals of 15 days between cycles, while co-trimoxazole (one DS tablet BD) is administered continuously for 35–105 days [1,8,9]. Ramam *et al.* initially described a two-step regimen consisting of an intensive phase with penicillin, gentamicin, and co-trimoxazole for 5–7 weeks, followed by maintenance therapy with amoxicillin and co-trimoxazole continued 5–6 months after clinical remission; however, they later modified this to gentamicin (1.5 mg/kg IV) plus co-trimoxazole (two double-strength tablets) given twice daily for four weeks followed by the continuation of co-trimoxazole plus doxycycline (100 mg twice daily) [8,9]. This modified approach had the advantage of reducing the number of injections and duration of the intensive phase and reducing the cost of therapy but still maintaining the efficacy [8,9]. We used the continuation phase of the modified Raman regimen of doxycycline and co-trimoxazole combination as the sole treatment since the patient was not ready for an extended period of admission for gentamycin injections. After three months of follow-up, the patient had complete regression of swelling. The same treatment has been continued for another 3–4 months. Thus, we further reduced the treatment cost for our patient and achieved a good outcome.

CONCLUSION

Mycetoma can have either fungal (eumycetoma) or bacterial (actinomycetoma and botryomycosis) etiology. The clinical presentation is usually a subcutaneous swelling with discharging sinuses and granules. The three types can be differentiated based on staining and bacterial and fungal cultures [1]. The extremities are the most common sites of involvement. However, actinomycetoma can present at unusual locations such as the scalp without the classic morphology [4,6,9]. It has to be kept in the differential diagnosis, especially in patients from endemic countries. Co-trimoxazole is the mainstay of treatment [1].

Conflict of interest: None declared.

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