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Case Report

Actinomycosis of the Foot: A Rare Case Report

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Abstract

Mycetoma is a chronic granulomatous disease affecting the skin and subcutaneous tissue. It tends to have a chronic course either caused by true fungi or filamentous bacteria. Current case report is a rare case of Mycetoma involving the foot due to actinomyces species. A 35-year-old male presented with 1 year history of multiple swellings over the left foot with discharging sinuses. Patient was treated with Cotrimoxazole and Amikacin combination and later treated with Doxycycline and Amikacin combination. This case report emphasis the importance of multidisciplinary approach in diagnosis and management of chronic granulomatous disease.

Keywords: Actinomycosis; Madura Foot; Subcutaneous Nodule

Case Report

A 35-year-old male farmer presented to a tertiary care orthopaedic centre with severe disability secondary to multiple wounds in his left foot which progressed slowly involving the entire foot. As described by patient he noticed a small swelling which was around size of a small marble after injuring himself with a stick while working in the farm. He developed multiple swellings along with purulent discharge from multiple sites (Figure 1). Patient had no history of fever, diabetes, varicosity, tuberculosis, or any other chronic conditions. The patient was treated with several oral and injectable antibiotics in the past one year before presenting to us.

At presentation, on physical examination there were multiple small ulcers with purulent discharge involving both dorsal and plantar aspect of his left foot. Also he had diffuse induration with change in texture of skin along with blackish pigmentation all over the foot. Vascularity was intact in the involved area of the foot. A provisional diagnosis of chronic osteomyelitis was made. Differential diagnosis of tuberculosis and chronic fungal infection was considered.

The aspirate from the lesion was sent to microbiology lab for Gram's staining, Ziehl Neelsen's staining, KOH wet mount, aerobic, anaerobic and fungal culture to get correct picture on the diagnosis of the condition. Also the tissue bits from the lesions were sent to pathology lab for histopathological examination. Gram's stain picture revealed numerous polymorphonuclear neutrophils and plenty of gram-positive filamentous bacilli. Ziehl Neelsen's stain was negative for acid fast bacill. KOH wet mount was negative for fungal filaments. In microbiology lab the specimen was inoculated on Blood agar, Mac Conkey agar and Thioglycolate broth and kept for incubation at 37°C. The specimen was also inoculated onto Blood agar and Robertson's cooked meat media and kept in McIntosh Filde's anaerobic jar for anaerobic culture. The specimen was also inoculated onto Sabouraud's Dextrose Agar (SDA) for fungal culture, one kept at incubation temperature at 37°C and the other kept at room temperature at 25°C.

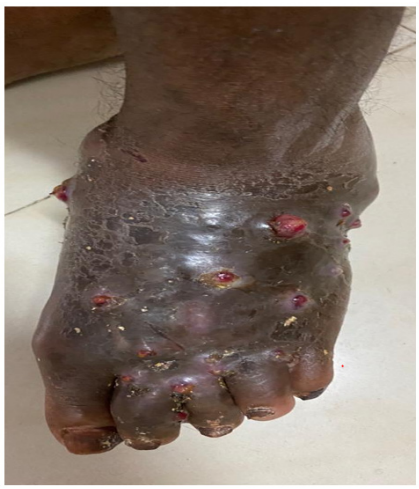

Figure 1: Multiple discharging sinuses on left foot at presentation.

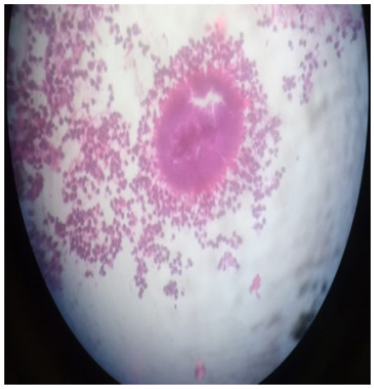

Figure 2: Histopathological examination showing actinomycotic colonies.

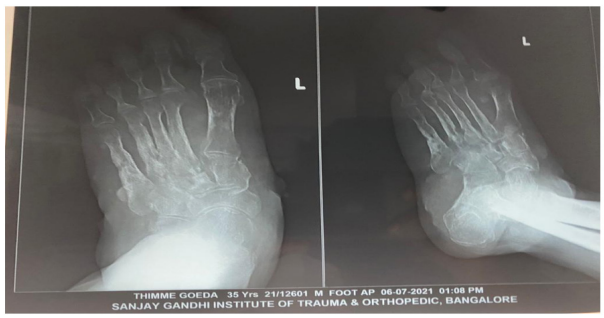

Figure 3: Foot showing sclerosis of bone with focal radiolucency.

Aerobic culture yielded no growth of organisms and anaerobic culture yielded scanty growth of white irregular colonies after 1 week of incubation on blood agar. The colony morphology was consistent with that of actinomyces species. There was no growth on SDA. The organism was identified as actinomyces species based on the morphological appearance on gram's stain, negative Modified ZN stain, growth on BA and other characters. Tissue samples collected from the lesions on Haematoxylin and Eosin stain showed multiple granulomas with dense network of thin gram-positive filaments surrounded by peripheral zone of club shaped structure presenting a sun ray appearance. Also histopathological examination revealed actinomycotic colonies with Splendore-Hoepple phenomenon along with suppurative granulomas composed of multinucleated giant cells, epitheliod cells, lymphocytes and neutrophils (Figure 2).

Mantoux test was negative. X-ray of left foot showed soft tissue swelling with patchy sclerosis of bones with focal lucencies and osteoporosis.

The patient was treated with combination of injectable Amikacin 500mg IV BD along with Co trimoxazole and Sulfamethaxazole 80mg + 400mg BD for 1 month followed by Doxycycline 100 mg BD with Cotrimaxazole DS for another 5 months. Following this, lesions healed completely with residual swelling and pigmentation, planned to follow up on OPD basis. The patient responded well to treatment and the lesions healed with scar formation.

Discussion

Mycetoma is classically described as "Madura foot" is characterised with a triad of subcutaneous swellings, formation of sinuses and discharge containing grains. It is a chronic, slowly progressive granulomatous subcutaneous infection. More than 20 species of filamentous bacteria are described to be causative agents [1].

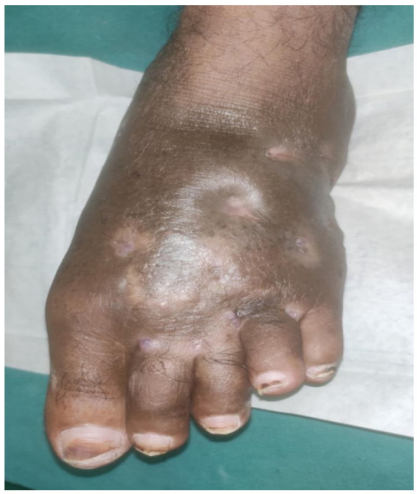

Figure 4: Healed wound with scarring after two months.

Mycetoma is either Eumycotic or Actinomycotic in aetiology. Important species causing Actinomycotic mycetoma belong to the genus Actinomadura, Nocardia and Streptomyces species [2].

Actinomycosis involving extremities is rare but has been reported in literature. Only few cases from an infection in the leg have been reported [3,4]. Three important clinical presentations should prompt this diagnosis : 1. the chronicity of a mass like lesion that extends through tissue borders and mimics a tumor, 2.the development of sinus tracts that spontaneously resolve and recur, and 3. a relapsing infection following a short course of antibiotics [5].

Actinomycosis has been called the "most misdiagnosed disease" and has been a diagnostic challenge even to most experienced clinicians. Its chronic and indolent course resembles that of tuberculosis, fungal infection, malignancy and delays early diagnosis of treatment [6]. This was the scenario in our case too where patient was misdiagnosed and was treated with several antibiotics before histopathology, Gram's stain smear, Zeihl Nielsen stain smear and culture results were available.

An appropriate diagnosis of actinomycosis requires a combination of microbiological, pathological and molecular studies. The clinical diagnosis starts with obtaining a sample of suppurative exudate, tissue or sulphur granules. Diagnosis of Actinomycosis is considered when a direct gram stain of the suppurative exudates or histologic section shows Gram positive, non-acid fast rods in diphtheroidal arrangements with or without branching [7].

Osteomyelitis due to actinomyces has been reported infrequently in adults [8]. Bone involvement occurred in 1-15% of those series reviewed by Lewis and associates [9]. Hematogenous spread of actinomyces with extra osseous granuloma formation and minimal sub periosteal bone reaction has been reported by Gholamreza R., *et al.* [10].

Differentiation between Actinomycetoma and Eumycetoma is Important as treatment options for both of them differ. A study suggested a modified two step treatment for actinomycetoma consisting of an intensive phase in which a combined treatment of intravenous gentamicin with oral cotrimoxazole twice daily is given for a period of 4 weeks followed by maintainance phase with cotrimoxazole and doxycycline given twice daily for 5-6 months [11]. Combination therapy is usually recommended for treatment of mycetomas. Early diagnosis and treatment is important to avoid amputations. Continuous clinical follow-up is needed to detect recurrence which is common.

Conclusion

Keeping the differential open and prompt local examination, detailed microbiological and histopathological examination helped to diagnosing this case of Madura foot which usually masquerades its presentation either due to chronicity or partial treatment.

Conflicts of Interest

The authors declares that there is no conflict of interest regarding the publication of this paper.

Funding Statement

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