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Nocardial Mycetoma pedis: A Case Report Study

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Abstract

Nocardiosis is an uncommon infection caused by *Nocardia* species, a group of aerobic actinomycetes. Nocardiosis may affect immunocompromised or immunocompetent patients and as this case was affected by a rare disorder, we describe it in this study. This paper describes a case of nocardial mycetoma from a hospital affiliated to Shiraz University of Medical Sciences. The case was a 52-year-old man with diabetes mellitus, who was admitted to our institution with a 5-year history of right lower extremity swelling with multiple discharging sinuses localized in distal part of the leg and dorsum of the foot following a penetrating injury to the affected foot. The wound culture yielded the growth of *Nocardia* species after incubation. Nocardiosis should be considered in the differential diagnosis of skin lesions, especially if patients have a history of trauma or contact with soil-contaminated materials. Our reported case increases knowledge about this rare, sporadic infection in our country.

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Introduction

Nocardiosis is a rare bacterial infection. *Nocardia* typically appears as filamentous gram-positive branching rods. In addition, most nocardiae are acid-fast in direct smears if a weak acid is used for decolorization (1). Cases of actinomycetoma had been reported in Fars province of Iran many years ago (2). Cutaneous nocardiosis (CN) may be classified into primary CN as a result of inoculation and secondary CN as a part of disseminated infection. Primary CN has three clinical variants: a superficial skin and soft tissue infection (cellulitis), lymphocutaneous type, and a deeper infection (nocardial mycetoma). Nocardial mycetoma is the most common type of primary CN (3). This infection usually involves the hands and feet. Mycetoma is a chronic cutaneous infection with an indolent course ranging from months to years that can be caused by actinomycetes (actinomycetoma) or fungi (eumycetoma). It forms small, subcutaneous swellings with

concurrent invasion of deeper tissues (1). Here, we present a case of primary CN (Nocardial Mycetoma) in a middle-aged man with diabetes mellitus.

Case Presentation

A 52-year-old Afghan man, resident of Shiraz, with a history of diabetes mellitus for unknown duration, was admitted to our institution with a 5-year history of right foot and leg swelling with multiple discharging sinuses localized in distal part of the right leg and dorsum of the right foot following a penetrating injury to the affected foot. The patient had been hospitalized three times during 5 years due to the skin lesions mentioned previously. There was no fever, chills or night sweats. The patient did not have any respiratory or urinary symptoms. Although he did not have a history of anorexia, he had a history of weight loss.

On admission, the patient was afebrile (36.6˚C orally), his pulse rate was 80 beats/min (BPM), respiratory rate was 16 breaths/min, and blood pressure was 110/60 mmHg. There were multiple nodular lesions in an edematous purplish base on distal part of the right leg and dorsum of the right foot. Multiple discharging sinuses draining purulent material with white granules were present (Figure 1A).

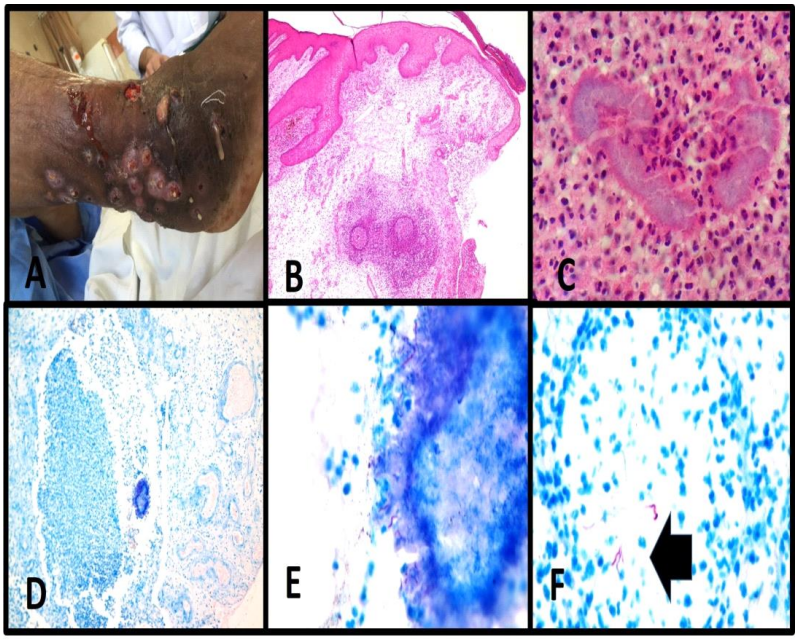

Figure 1. Clinical and histopathology picture. (A) Clinical picture of the infected foot with discharging sinuses and ulcers. (B,C) Hematoxylin and Eosin stained histopathology section of the skin lesions shows subepidermal and dermal inflammatory cell infiltration with microabscess formation (X100) around the basophilic filamentous and granular aggregates (C. sulfur granules- X400). (D,E,F) Acid-fast stained skin biopsy shows the mentioned granules containing branching, filamentous, partially stained bacteria representing of Nocardia spp. (Arrow) (X100, X400, X1000-oil-immersed).

On palpation, warmth and tenderness of the lesions were notable. Pain on motion and decreased range of motion of the right ankle were present. Distal pulses were symmetrically detectable. Rest of the physical examination was within normal limits. Laboratory data revealed mild leukocytosis with 86.6% neutrophils and severe normochromic normocytic anemia. Renal and liver function tests showed normal values. C-reactive protein (CRP) level and erythrocyte sedimentation rate (ESR)

were 192 mg/L and 110 mm/h, respectively. HIV, HCV, and HBV tests were negative. Insulin was started but was discontinued due to hypoglycemia, because during hospital course blood glucose profile was within the accepted range without using insulin or oral antihyperglycemic agents. The wound culture was done, then, intravenous trimethoprim-sulfamethoxazole (TMP-SMZ) was administered empirically. After consultation with an infectious disease specialist, Penicillin G- 4 million U (four times a day) was administered 10 days later. The soft tissue ultrasound showed soft tissue edema and formation of collection in contact with bone from the distal third of tibia to the foot. The MRI of ankle joint revealed cellulitis and diffuse osteomyelitis. Dot-in-circle sign was also present, which is highly indicative of mycetoma (Figure 2).

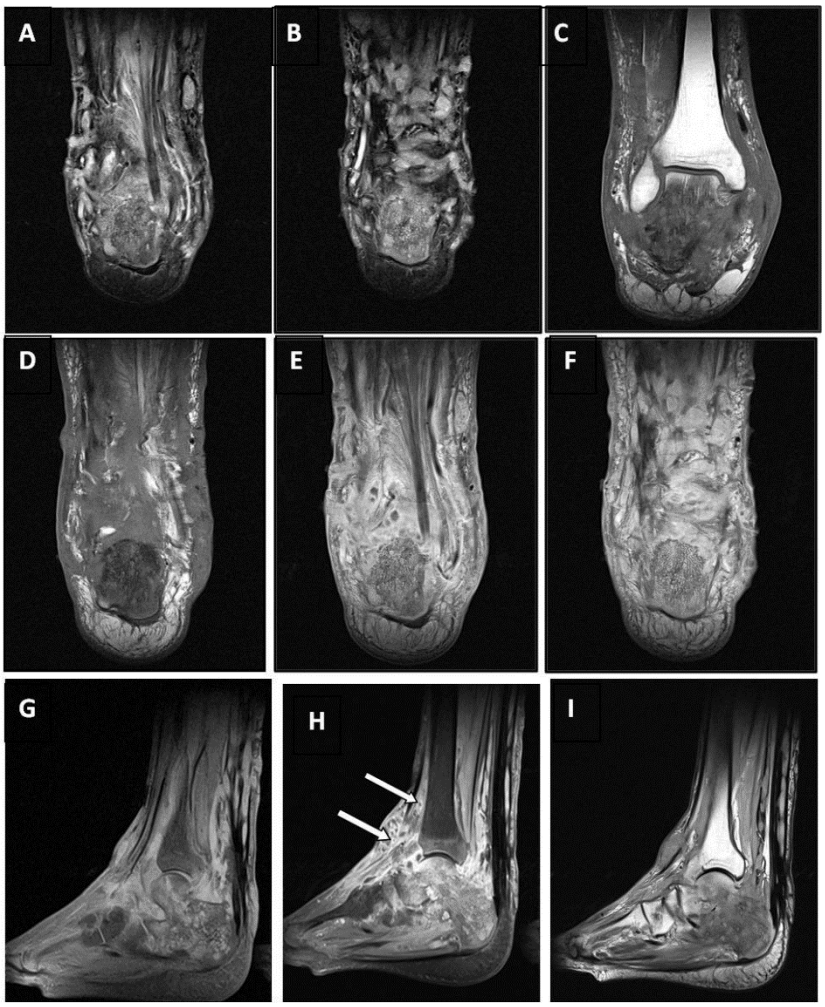

Figure 2. Radiology picture. (A, B) Short-TI inversion recovery (STIR) images in coronal plane that show multiple subcutaneous abscesses with fistula on the skin surface and osteomyelitis in calcaneus. (C, D) Non-contrast-enhanced T1-weighted images in coronal plane that show the abscesses. (E, F) Fat-saturated T1-weighted images after injection of contrast agent that reveal enhancement of subcutaneous abscesses and surrounding soft tissue. (G, H, I) Fat-saturated T1-weighted, STIR, and T2-weighted images in sagittal plane that reveal the same findings with clear involvement of talus and calcaneus. (H) Dot-in-circle sign (Arrows). The dot represents the central grains, and the circle represents the surrounding granulation tissue.

The skin biopsy showed many microabscesses in the upper dermis, which contained sulfur granules giving the impression of actinomycosis (Figures 1B and C). Acid-fast staining revealed partial acid fastness of filamentous organisms in the microabscesses giving the impression of nocardiosis (Figures 1D-F). The wound culture showed the growth of Nocardia four weeks after the start of incubation. Nocardia infection (nocardial mycetoma) was diagnosed. But we could not identify the exact type of involved Nocardia species due to the limitations of our lab facilities.

The patient was hospitalized for 33 days. Following antibiotic therapy, clinical improvement occurred. His last white blood cell (WBC) count was within normal range. Also, CRP level and ESR reached 24 mg/L and 102 mm/h, respectively. The patient was discharged with oral TMP-SMX and Penicillin V. Clinical cure occurred after 3 months but the use of drug continued for a further 6 months.

Discussion

Nocardiosis is a rare, opportunistic bacterial infection caused by *Nocardia* species which belong to a large group of bacteria, aerobic actinomycetes. Primary CN is more frequently seen among those with intact immune function through direct inoculation (4). Inamadar et al. (2003) have reported 10 cases of primary CN seen in a tertiary care hospital in South India over a period of ten years. All the patients had a history of preceding injury to the affected area and were agricultural workers or laborers, except for a house-wife. The most common clinical presentation was mycetoma (5 cases). An acute abscess was seen in 3 cases and a lymphocutaneous infection in 2 cases. Culture was positive in 6 cases. None of the patients had systemic involvement. All the patients had complete clinical resolution in 2-3 months with oral TMP-SMX, but treatment continued for 6 months (3). Although nocardial mycetoma is often localized to distal part of extremities, Seol et al. (2013) have reported a case of disseminated nocardial mycetoma in a patient who was on long-term corticosteroid therapy for the treatment of microscopic polyangiitis. The patient presented tender erythematous nodules scattered on the scalp, arm, leg, and abdomen (5).

Diagnosis of suspected CN requires examination of draining plus for curved, branching, beaded, gram-positive filaments. Granules which are macroscopic colored grains are frequently found in discharges of lesions in actinomycetoma but almost never in other forms of nocardiosis. Most nocardiae are acid-fast in direct smears if modified acid-fast stain is used for demonstrating these microorganisms. They are slow-growing and it may take a few weeks for them to be isolated from a clinical specimen (1). MR imaging is the modality of choice to depict soft tissue and bone involvement. At MR imaging, musculoskeletal mycetomas are characterized as conglomerates of multiple small rounded T2 hyperintense areas of granulation tissue, with central low-signal-intensity grains. The dot-in-circle sign was first described by Sarris et al. (2003) in two cases of soft tissue mycetoma of the foot. This sign is appreciated on T2-weighted, proton-density, short inversion time inversion-recovery, and gadolinium-enhanced T1-weighted fat-saturated images. It is highly indicative of mycetoma and is seen in 80% of patients (6). Patients with primary CN respond very well to medical therapy. Clinical response to the treatment with TMP-SMX was evident within a few weeks, but the use of drug has to be continued for a further

several months (6-12 months after clinical cure in actinomycetoma) to prevent recurrence (1).

Conclusion

Our reported case increases knowledge about this rare and sporadic infection, Nocardiosis, in our country. Nocardiosis should be considered in the differential diagnosis of acute or chronic skin lesions especially if the patient has a history of preceding trauma or contact with soil-contaminated material. Delay in diagnosis of the responsible pathogen may lead to significant morbidity and deformity of the affected extremity, as presented in our patient.

References

1. Filice GA. Nocardiosis. In: Jameson JL, Fauci AS, Kasper DL, Hauser SL, Longo DL, Loscalzo J. Harrison's Principles of Internal Medicine. 20th ed. USA: McGraw-Hill; 2018.
2. Griffiths WA, Kohout E, Vessal K. Mycetoma in Iran. International Journal of Dermatology 1975; 14(3):209-13.
3. Inamadar AC, Palit A. Primary cutaneous nocardiosis: a case study and review. Indian J Dermatol Venereol Leprol 2003; 69(6):386-91.
4. Ambrosioni J, Lew D, Garbino J. Nocardiosis: updated clinical review and experience at a tertiary center. Infection 2010; 38(2):89-97.
5. Seol CA, Sung H, Kim DH, Ji M, Chong YP, Kim MN. The first korean case of disseminated mycetoma caused by nocardia pseudobrasiliensis in a patient on long-term corticosteroid therapy for the treatment of microscopic polyangiitis. Ann Lab Med 2013; 33(3):203-7.
6. Sarris I, Berendt AR, Athanasous N, Ostlere SJ; OSIRIS collaborative study group. MRI of mycetoma of the foot: two cases demonstrating the dot-in-circle sign. Skeletal Radiol 2003; 32(3):179-83.