

Case 20

A 62-year-old Thai female from Mukdahan

Chief complaint: Gradually enlarged indurated plaque on dorsum of right foot for 13 years



Fig. 20.1

Present illness:

The patient noticed swelling on the dorsum of her right foot that gradually enlarged for 13 years. Multiple small ulcers developed on the surface (Fig. 20.1). Skin biopsy was done and revealed suppurative granuloma with organism colony and positive PAS stain. She was diagnosed with eumycotic mycetoma complicated with osteomyelitis and had been treated as mycetoma for 2 months without improvement. Thus, patient came to re-visit dermatology out-patient clinic.

Past history: She had type-2 diabetes mellitus, essential hypertension, dyslipidemia and chronic kidney disease for 17 years. Her current medication includes glipizide, metformin,

simvastatin, CaCO_3 , NaHCO_3 , folic acid, ferrous fumarate. Her occupation is farmer.

Physical examination:

- Except for tenderness on weight bearing at right foot, other physical examination revealed no abnormalities.

Dermatological examination:

- Swelling, indurated, subcutaneous plaque overlying with multiple draining sinuses on right foot.

Histopathology: (S19-000302, Right foot)

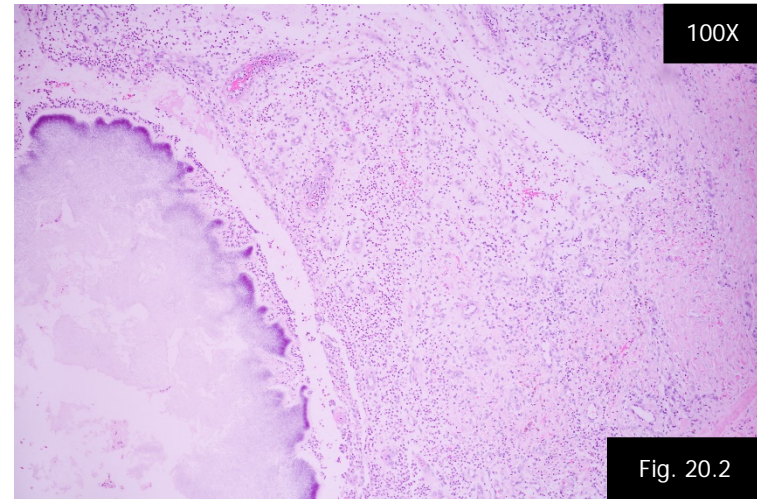


Fig. 20.2

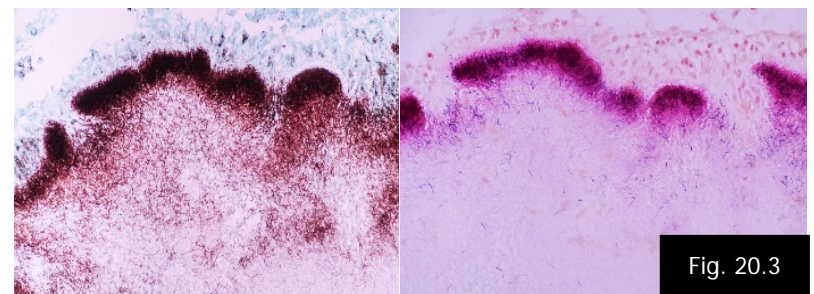


Fig. 20.3

- Chronic abscess with numerous neutrophils and fibrosing granulation tissue (Fig. 20.2)
- Grains showing beaded gram positive filamentous bacteria highlighted by Brown-Brenn and GMS staining (Fig. 20.3)
- Negative AFB and Fite staining

Laboratory investigations:

- CBC: WBC 6700 /cumm, PMN 59%, Lymphocyte 33%, Monocyte 4%, Eosinophil 3%, Basophil 1%, Hb 8.6 g/dL, Hct 28.8%, MCV 74.2 fL, Platelets 314,000 /cumm
- BUN 20 mg/dL, Cr 1.49 mg/dL, AST 16 U/L, ALT 11 U/L, ALP 98 U/L, GGT 14 U/L, Albumin 34.4 g/L, Total protein 86.1 g/L, Total/Direct bilirubin 0.4/0.2 mg/dL
- Tissue microbiology
 - Aerobe culture: no growth
 - Fungal culture: no growth
- Film right foot: Osteolytic lesions

Diagnosis: Actinomycetoma with chronic osteomyelitis

Treatment:

- Amikacin 500 mg intravenous once daily then switch to oral amoxicillin/clavulanate 375 mg 1 tab per oral twice daily (renal dosage adjustment)
- Sulfamethoxazole/Trimethoprim 400/80 mg 3 tabs per oral in the morning and 2 tabs in the evening.

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Discussion:

Mycetoma or Madura foot is a chronic suppurative infection of the skin, subcutaneous tissue and bones. Mycetoma-like condition was first described since 1842 in Madurai, India.¹ Mycetoma is caused

by either fungi or bacteria, and classified as eumycetoma and actinomycetoma, respectively. It occurs more common in men and the predilection site is foot. However, other affected sites including head, scalp, shoulders, neck, trunk, arms, breast, back, buttocks, elbows, palms, groins, vulva, thighs and knee were also reported in literature.²

After inoculation, actinomycetoma gradually develops. Hemispherical swellings of affected extremities is among the first symptoms. The skin is not warm and painless firstly. They become more painful due to secondary bacterial infection and infection spreading to other underlying structures like osteomyelitis. The typical clinical features is a triad of localized swelling, draining sinus tracts and producing of grains or granules. Fever is uncommon.³

Nocardia, *Streptomyces* and *Actinomadura* were among most common pathogens, depending on each regions.¹ Characteristics and color of grains are useful to differentiate between causative organisms. Pale white to yellow grains are from *Actinomadura madurae* and *Nocardia* spp. While yellow-brown grains are from *Streptomyces* spp. and pink-red grains are from *Actinomadura pelletierii*. On the other hand, eumycetoma mostly produces black grains, except for *Pseudallescheria boydii*, *Fusarium* spp., *Acremonium* spp. and *Aspergillus* spp.^{1,4} Histopathological examination and microbiological evidence are helped in making diagnosis. Actinomycetoma also shows a Splendore-Hoeppli reaction, eosinophilic and pseudomycotic structures composed of necrotic debris and immunoglobulins form rings around the grains.⁵ However, grains produced from different organisms also show different characteristics as shown in Table 20.1. If actinomycetoma is suspected, gram stain and Brown-Brenn stain should be performed. Selective media like Lowenstein-Jensen media or Sabouraud dextrose agar are commonly used in

actinomycetoma isolation. Imaging should be done to surveillance for bones and joints involvement.⁶

For treatment, amikacin (15mg/kg/day) for 3 weeks and trimethoprim-sulfamethoxazole (TMP-SMX, 40/8mg/kg/day every 12 hours) for 5 weeks are the first-line treatment of actinomycetoma. Amoxicillin-clavulanate is alternatively used in resistant cases.⁶ Some other antibiotics including rifampicin, imipenem, carbapenem and oxazolidinones were used in some case reports.³ Further surgical treatment and amputation are spared for resistant cases or patients with severe bone destruction.⁶

In our case, patient was suspected to have Nocardial mycetoma due to presence of filamentous colonies in Brown-Brenn, GMS and PAS stain. *Nocardia* spp. can be stained with gram stain, modified Kinyoun stain, Ziehl-Nielsen stain, and Brown-Brenn stain.⁷ Due to osteomyelitis, orthopedics consultation was done. Patient was advised to wear hard sole shoes to prevent pathologic fractures. After treatment, swelling and grains gradually subsided.

Table 20.1 Distinction between actinomycetoma and eumycetoma

	Actinomycetoma	Eumycetoma
Staining	Gram-stain or Brown-Brenn stain	GMS or PAS stain
Size of organism	0.5-1 µm-wide filaments	2-5 µm-wide hyphae
Characteristic of organism	Septate fine-branching filaments	Septate hyphae

GMS; Gomorimethamine silver, PAS; Periodic acid-Schiff

Adapted from Relhan V, Mahajan K, Agarwal P, *et al.* Mycetoma: An Update. *Indian J Dermatol.* 2017;62(4):332-40.

References:

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