Case 8

Draining mass on the ankle

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Patient: A 48-year-old Thai farmer woman from Chaiyaphum

Chief complaint: Mass on right ankle for 18 years

Present illness:

The patient has developed a mass on right ankle for 18 years. This lesion began with a small painless nodule with gradually enlarged. She noticed that the lesion showed intermittent yellowish discharge with white grains. Her right foot became swollen and painful when she walked.

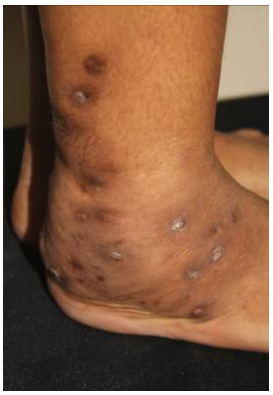
Past history: No known underlying disease

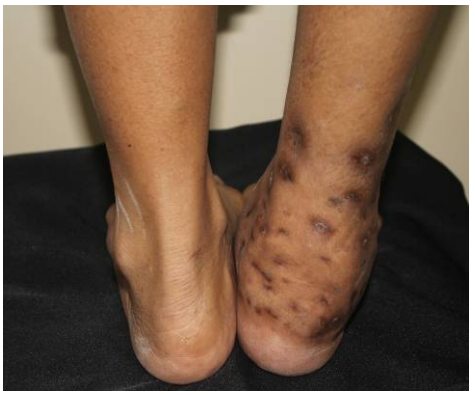
Skin examination:

- A hyperpigmented mass with coalescing nodules with sinus tracts over the surface on right ankle
- Yellowish discharge and white grains are seen
- Right ankle is marked swelling and tender

Histopathology: (S13-17619, right ankle)

There is scatter fibroblast, new-formed collagen bundles and increase number of thick-wall blood vessels in association with sparse perivascular inflammatory cell infiltration.





Investigation:

Microbiology tests

Gram stain: Gram positive filamentous bacteria
AFB and mAFB stains: Negative
Culture for aerobe: No growth
Culture for fungus: No growth
16S and 18S ribosomal RNA sequencing: Negative

Imaging studies

Plain film of the right ankle:

- Multiple osteolytic lesions at the right calcaneus associated with soft tissue swelling of right foot, which could be osteomyelitis

MRI of the right ankle:

- Chronic osteomyelitis of right talus and calcaneus as well as evidence of mild surrounding myositis
- Mild arthritis bone changes of ankle with tenosynovitis of tibialis posterior and flexor hallucis longus and brevis, represent chronic inflammation process

Diagnosis: Mycetoma with osteomyelitis of the right ankle

Treatment:

- Total 5 cycles of
  - Amikacin 500 mg iv q 12 hr (3 week/cycle)
  - Co-trimoxazole (80/400) 2 tabs oral bid (5 week/cycle)

Discussion:

Mycetoma is a chronic granulomatous subcutaneous slowly progressive infections in which true fungi (Eumycetes) or filamentous bacteria (Actinomycetes),[1] generate grain-filled pus via multiple draining sinuses onto skin or involve adjacant bone, causing osteomyelitis.[2]

Both forms of mycetoma present as a progressive, subcutaneous swelling, although actinomycetoma has a more rapid course. The legs are the most commonly affected site, but actinomycetoma can be presented in most regions of the body.[3] Mycetoma is usually painless suggested that the mycetoma produces substances that have an anaesthetic action. Pain may be produced by the expansion of the bone. Skin changes are common in mycetoma. Regional lymphadenopathy is not uncommon. Cachexia and anemia may be seen in late mycetoma. This is often due to malnutrition, sepsis and mental depression. Mycetoma can produce many disabilities, distortion and deformity.[4]

This disease is endemic in tropical, subtropical, and temperate regions of the world. 60% of mycetomas are caused by actinomycetes, although the incidence can vary regionally due to geographic variations. Mycetoma predominates in men for all causal agents with the exception of Actinomadura madurae that favors women.[4] It occurs between the fourth and fifth decades of life. It is interesting that during pregnancy and children, mycetoma becomes more active and aggressive that may cause from changing in hormonal environment and decrease immune response. The role of the immune system in mycetoma has been studied and believed that a Th2-response might be necessary to induce mycetoma.[5]

This disease has been associated with minor trauma caused by thorn pricks, stones, snake or insect bites. Identifying agent is essential because treatment varies completely in each type of infection.[6]

Diagnosis may involve radiology, ultrasonic imaging, cytology, culture, histology or immunodiagnosis.[7] The culture technique is often cumbersome, time consuming and contamination may give a false positive result. Direct microscopic examination of the pus with 10% KOH or saline reveals the presence of granules. Their size, form, and color can identify the probable causative agent. Histopathology of a deep surgical biopsy is always required to

confirm the diagnosis.[5]

Radiography shows a dense shadow of soft-tissue granuloma and calcification. The cortex may be compressed leading to scalloping and punched-out cavities. MRI is helpful in assessing the extent of bone destruction, periosteal reaction and soft-tissue involvement. High signal intensity within granuloma give "a dot-in-circle appearance" that indicates the presence of grains and is highly specific for mycetoma.[5,8,9]

The differential diagnosis of mycetoma includes sporotrichosis, tuberculosis, osteomyelitis, coccidiomycosis, phaeohyphomycosis, botryomycosis, other fungal infections, and neoplasias of the bone and soft tissues.[3]

Diagnosis of the causal agents of mycetoma is a prerequisite for proper treatment. The current therapy includes medical treatment or a combination of medical treatment and surgery.[10]

The treatment of choice for uncomplicated actinomycetoma is trimethoprim-sulfamethoxazole for 6 months to 2 years. If not response, amoxicillin-clavulanic acid can be used for 6 months. In complicated case, combined treatment with amikacin (15 mg/kg/day) in a divided dose every 12 hours for 3 weeks and trimethoprim-sulfamethoxazole (8/40 mg/kg/day) for 5 weeks for a period of 5 to 20 weeks is indicated.[5] In our patient, the clinical improved after full course of combined antibiotics.

References:

1. Fahal AH. Mycetoma: a thorn in the fleshTrans R Soc Trop Med Hyg. 2004; 98(1): 3-11.
2. Fahal AH, Shaheen S, Jones DH. The orthopaedic aspects of mycetoma. Bone Joint J. 2014; 96-B(3): 420-5.
3. Hogade S, Metgud SC, Swoorooparani. Actinomycetes mycetoma. J Lab Physicians. 2011; 3(1): 43-5.
4. Dlova NC, Mosam A. Actinomycetoma. Skinmed. 2012; 10(2): 98.
5. Welsh O, Vera-Cabrera L, Welsh E, Salinas MC. Actinomycetoma and advances in its treatment. Clin Dermatol. 2012; 30(4): 372-81.
6. Mattioni S, Develoux M, Brun S, et al. Management of mycetomas in France. Med Mal Infect. 2013; 43(7): 286-94.
7. Chufal SS, Thapliyal NC, Gupta MK. An approach to histology-based diagnosis and treatment of Madura foot. J Infect Dev Ctries. 2012; 6(9): 684-8.
8. Bouziane M, Amriss O, Kadiri R, Adil A. The role of computed tomography in the exploration of Madura foot (pedal mycetoma). Diagn Interv Imaging. 2012; 93(11): 884-6.
9. Cherian RS, Betty M, Manipadam MT, et al. The "dot-in-circle" sign -- a characteristic MRI finding in mycetoma foot: a report of three cases. Br J Radiol. 2009; 82(980): 662-5.
10. Agarwal US, Besarwal RK, Gupta R, Agarwal P. Treatment of actinomycetoma foot--our experience with ten patientsJ Eur Acad Dermatol Venereol. 2013; 27(12): 1505-13.