

Case Report**Cervical cord compression secondary to mycetoma infection**

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Abstract

Mycetoma is a chronic granulomatous subcutaneous infection caused by actinomycetes (actinomycetoma) or by true fungi (eumycetoma). Clinically the disease is characterized by swelling, and sinuses in the affected part. Another characteristic feature of mycetoma is the formation of aggregates of the organism (grains) in the tissues, which are visible to the naked eye and are discharged through sinuses in the skin. The grains vary in colour, size and consistency depending on the causative agent^(1,2). These features are helpful in making a tentative diagnosis of the causative organism. We report a 75 year's Sudanese female with spastic paraplegia of gradual onset, she has discharging sinuses in her neck. Cervical X-RAY and cervical MRI showed destruction of cervical vertebrae. Identification of the agent was established by the histological examination of the grains, by culture and serologic techniques. Mycetoma is a local chronic and progressive infection of the skin, subcutaneous tissues and bone. It is characterized by swelling that is often grotesque and disfiguring and by multiple sinus tracts that drain granule-containing pus⁽³⁾.

Introduction

Mycetoma is caused by at least 20 species of actinomycetes and fungi. The most common infecting agents are *Nocardia* spp and *Madurella mycetomi*. The disease is acquired by traumatic implantation in the skin. The microorganisms grow through the subcutaneous tissue into the bone. As this occurs, there is hyperplasia of the tissues, formation of pus containing granules, and expression of pus to the surface of the skin and granuloma formation at the periphery of the infected area. The classical triad of symptoms is: Gross deformity of the infected area, draining sinuses and granules in the pus. Diagnosis is made by the presence of the classical triad of symptoms and by culture of the pus draining from the wound. Treatment is initially with trimethoprim-sulfamethoxazole for *Nocardia* or Amphotericin B

for fungi. If the fungal infection does not respond to amphotericin B then amputation is required⁽⁴⁾.

Case report

A seventy-five year's Sudanese female presented with upper and lower limbs weakness of gradual onset. First it affected the upper limbs and after three months both lower limbs were affected until she became unable to walk. This was not associated with numbness or sphincteric disturbance. She has no symptoms referable to cranial nerves involvement. She has no headache, convulsions, or loss of consciousness. The condition was preceded by febrile illness and the patient noticed skin pigmentation on the neck and it was not associated with itching. This was followed by multiple discharging sinuses on both sides of the neck. The grains expressed through the sinuses were yellow and white in colour. Some of the sinuses closed

transiently after discharge. The discharge is serosanguinous. First the condition was painless then the patient experienced neck pain. She has no symptoms referable to the cardiovascular, respiratory, urinary or musculoskeletal systems. She has no past history of diabetes mellitus or hypertension. She has no family history of similar condition. She has four children, her husband died before three years. She lives in her own house in JABAL AWLIA, twenty kilometers to the South of Khartoum. She doesn't use to smoke or consume alcohol. She became unable to cope with her normal domestic activities. Clinical examination revealed an ill cachexic pale patient, not jaundiced or cyanosed. Pulse 76/m. BP 120/85. Tem. 37°C. She is fully conscious, oriented in time, person and place. Both recent and remote memories are intact. She is cooperative. Cranial nerves are intact. She has weakness of neck flexion. Both upper limbs showed distal wasting, there is no deformity, no abnormal position, no abnormal movement and no fasciculation. She has bilateral hypertonia and hyperreflexia. Power is grade three proximally and grade two distally. All modalities of sensations are intact including light touch, pin prick, vibration and position. She has normal coordination. Examination of the lower limbs showed generalized wasting but there is no deformity, abnormal position or fasciculation. She has bilateral hypertonia and hyperreflexia. Power is grade three proximally and grade two distally. All modalities of sensations are intact including light touch, pin prick, vibration and position. She has normal coordination. Examinations of the back revealed no abnormalities. Local examination of the neck showed that the skin over it becomes attached and stretched. Also it became smooth, shiny and there are areas of hypo and hyper-pigmentation. There are subcutaneous masses with discharging sinuses some of the old ones were healed completely. There

is area of local hyperhidrosis confined only to the site of the lesion and the skin around it. The swelling is painless firm rounded, lobulated and mobile. She has cervical lymphadenopathy. A list of investigations were done including urine and stool analysis, complete haemogram, blood urea and electrolyte and liver function test, all were normal. Cervical X-ray showed an evidence of soft tissues swelling and destruction of cervical vertebrae C4 and C5. Cervical MRI showed extensive neck pyomyositis, epidural extension and cord compression plus focal cord infection at the medullary cervical region – appearance of chronic infection and goes with madura. The primary symptoms of a tumor, sinuses, and grain-flecked discharge provided enough information to diagnose mycetoma. The species of fungi at the root of the infection is identified by staining the discharge grains and inspecting them with a microscope.

Discussion:

Mycetoma has a worldwide distribution but this is extremely uneven. It is endemic in tropical and subtropical regions. The African continent seems to have the highest prevalence⁽⁵⁾. It prevails in what is known as the mycetoma belt stretching between the latitudes of 15 south and 30 north. The organisms are usually present in the soil in the form of grains. The infecting agent is implanted into the host tissue through a breach in the skin produced by trauma caused by sharp objects such as thorn pricks, stone or splinters⁽⁶⁻⁷⁾. The commonest site for mycetoma is the foot, most of the lesions are seen on the dorsal aspect of the forefoot. The hand ranks as the second commonest site. In endemic areas other parts of the body may be involved but less frequently and these include the knee, arm, leg, head and neck, thigh and the perineum. Rare sites such as the chest, abdominal walls, facial bones, mandible, paranasal sinuses, eyelid, vulva, orbit, scrotum and surgical incisions may be affected. The

clinical presentation of mycetoma is almost identical irrespective of the causal organism⁽⁸⁾. However, the rate of progress is more rapid with actinomycetoma than with eumycetoma. In eumycetoma, the lesion grows slowly with clear defined margins and remains encapsulated for a long period, whereas, in actinomycetoma the lesion is more inflammatory, more destructive and invades the bone at an earlier period⁽⁹⁾. The characteristic triad, of a progressive painless subcutaneous swelling at the site of previous trauma as well, the nodules may suppurate and drain through multiple sinus tracts and these sinuses may close transiently after discharge during the active phase of the disease. Fresh adjacent sinuses may open while THEY are connected with each other, with deep sterile abscesses and with the skin surface. The discharge is usually serous, serosanguinous or purulent. During the active phase of the disease the sinuses discharge grains (fungal colonies). Mycetoma is usually painless in nature, it was suggested that the mycetoma produces substances that have an anaesthetic action⁽¹⁰⁻¹¹⁾. At a late stage of the disease the pain may become negligible due to nerve damage by the tense fibrous tissue reaction, endarteritis obliterans or poor vascularisation of the nerve. Pain may be produced by the expansion of the bone with the mycetoma granuloma and grains or it may be due to secondary bacterial infection. As the mycetoma granuloma increases in subcutaneous tissue, fat, muscles and bone. For unknown reasons, the tendons and the nerves are curiously spared until very late in the disease process, this may explain the rarity of neurological and trophic changes even in patients with long standing mycetoma⁽¹²⁻¹³⁾. The absence of trophic changes may also be explained by the adequate blood supply in the mycetoma area. In the majority of patients the regional lymph nodes are small and shotty⁽¹⁴⁾. An enlarged regional lymph

node is not uncommon and this may be due to secondary bacterial infection, genuine lymphatic spread of mycetoma or it may be due to immune complex deposition as part of a local immune response to mycetoma infection. Mycetoma can produce many disabilities, distortion and deformity⁽¹⁵⁻¹⁶⁾. It can be fatal especially with cranial mycetoma.

Figure 1: Discharging sinuses



Figure 2: MRI cervical spine showing extensive neck pyomyositis, epidural extension and cord compression plus focal cord infection at the medullary cervical region – appearance of chronic infection and goes with Madura



References

- 1- Abbott PH. Mycetoma in the Sudan. *Transactions of the Royal Society of Tropical Medicine and Hygiene*. 1956; 50(1): 11-24.

- 2- Mahgoub ES and Murray IG. *Mycetoma*. London: William Heinemann, Medical Books Ltd. 1973, pp. 1-50.
- 3- Gonzalez-Ochoa A. Mycetoma. In: Canizares O, editor. *Clinical Tropical Dermatology*. Oxford: Blackwell Scientific; 1975. p. 24-29.
- 4- Magana M. Mycetoma. *International Journal of Dermatology*. 1984; 23(4): 221-236.
- 5- Fahal AH and Hassan MA, Mycetoma. *British Journal of Surgery*. 1992; 79(11): 1138-1141.
- 6- Fahal AH and Suliman SH. The clinical presentation of mycetoma. *Sudan Medical Journal*. 1994; 32: 46-65.
- 7- Fahal AH, EL Sheik H and EL Hassan AM. Pathological fracture in mycetoma. *Transactions of Tropical Medicine and Hygiene*. 1996; 90(6): 675-676.
- 8- Fahal AH, EL Hassan AM, Abella AO and Sheik HE. Cystic mycetoma: An unusual clinical presentation of *Madurella mycetomatis*. *Transactions of the Royal Society of Tropical Medicine and Hygiene*. 1998; 92(1): 66-67.
- 9- Fahal AH, Yagi HI and EL Hassan AM. Mycetoma-induced palatal deficiency and pharyngeal plexus dysfunction. *Transactions of Tropical Medicine and Hygiene*. 1996; 90(6): 676-677
- 10-Gumaa SA, Mahgoub ES and EL Sid MA. Mycetoma of the head and neck. *American Journal of Tropical Medicine and Hygiene*. 1986; 35(3): 594-600.
- 11-EL Hassan AM and Mahgoub ES. Lymph nodes involvement in mycetoma. *Transaction of the Royal Society of Tropical Medicine and Hygiene*. 1972; 66(1): 165-169.
- 12-Fahal AH, EL Sheikh HA, Homeida MA, EL Arabi YE, and Mahgoub ES. Ultrasonographic imaging of mycetoma. *British Journal of Surgery* 1997; 84(8): 1120-1122.
- 13-EL Hag IA, Fahal AH and Khalil EAG, Fine needle aspiration cytology of mycetoma. *Acta Cytologica*. 1996; 40(3):461-464.
- 14-Mahgoub ES. Mycetoma. *Seminars of Dermatology*. 1985; 4: 230-239.
- 15-EL Hassan AM, Fahal AH, EL Hag IA and Khalil EAG. The pathology of mycetoma: Light microscopic and ultrastructural features. *Sudan Medial Journal*. 1994; 32: 23-54.
- 16-Gumaa SA, Mahgoub ES. Evaluation of the complement fixation test in the diagnosis of Actinomycetoma. *Journal of Tropical Medicine and Hygiene*. 1973; 76(6): 140-142.