

Case report

Report of two cases of mycetoma in the Islamic Republic of Iran

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Introduction

Mycetoma is a chronic fungal disease of the skin and subcutaneous tissues which often invades the muscle fascia and bones [1–3]. It is characterized by the formation of sinuses and fistulous tracts that discharge sero-purulent exudates containing “sulfur granules” [4].

Distribution of mycetoma is worldwide, but it is largely confined to tropical climates. [5,6]. It is common in Africa, Mexico, Central America, India, some South American countries and certain regions of the eastern areas of the Middle East. A few cases of this disease have been reported in parts of the Islamic Republic of Iran such as Ahwaz, a south-western province. Up till 1975, 7 cases had been reported in the Islamic Republic of Iran; later 5 more cases were reported [7–9]. This is a report of 2 more cases from Kerman, a south-eastern province, bringing the total number of cases reported in the last 25 years to 14.

Case reports

Case 1

A 41-year-old man from Kahnooj, a city in Kerman province, was referred to the Ker-

man Darman dermatology clinic in the year 2000 with a chief complaint of a painless swollen plaque on the back of his right foot which had appeared 35 years before. It had started with a 4.5 × 4.5 cm dark, firm nodular lesion that enlarged after a few months and draining sinuses formed above it. The lesion had become painful with time, so he had some difficulty in walking. Later radiographic examinations revealed destructive extensions to the deep tissues, especially to the metatarsus bone. Because of this and the pathology report, osteomyelitis had been diagnosed and he was treated. He had relative improvement for 10 years but then, following a trauma to his foot, the lesion became swollen and painful with draining sinus tracts. Surgical treatment had been carried out at his last examination but, despite drainage, the swelling increased and discharging ulcers appeared as nodulocystic lesions. As a result he could not walk without a walking stick. He was admitted to Kerman Darman hospital but nothing was found on general examination except weight loss and positivity for rheumatoid arthritis and C-reactive protein. Radiographic examination showed osteolytic lesions in the metatarsal bones to the point that complete destruction of a second metatarsus was seen. The pathology

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results of excised skin section showed vesicle formation and spongiosis, with chronic and acute inflammatory cells; infiltration cells included many plasma cells and some neutrophils through the dermis and subcutis. Excision biopsy of deep cut sections of the lesion revealed sulfur granules with a positive periodic acid Schiff reaction and eumycotic mycetoma was suggested. Combination of oral trimethoprim and sulfamethoxazole (co-trimoxazole) 800 mg twice daily was administered. After 1 month of therapy and management of lesions for 2 months, the pain disappeared, swelling was reduced and obvious healing was observed.

Case 2

A 38-year-old man developed pain and swelling on his right ankle for 1 year after suffering trauma. He attended Kerman Darman hospital in 1999 and was diagnosed with infective arthritis and was given treatment but there was no improvement after 6 months; ankle pain and difficulty in walking added to his problem. The patient was referred to the dermatology clinic of Kerman Darman hospital and, on physical and clinical examination, a hard mass was found on in his ankle that caused restriction in joint movement. On general examination, no abnormality was found except weight loss. Radiographic examination revealed an increase in soft tissue density. Fine-needle aspiration of the swollen area was performed and the laboratory result showed dark-coloured grains; the diagnosis of eumycetoma was thus made. Excision biopsy of cut skin showed severe chronic inflammation and induration in association with acute inflammatory cell infiltration, which included many plasma cells and some neutrophils in the dermis and also the subcutis. A trial therapy was attempted with a combination of trimethoprim and sulfamethoxazole (co-trimoxazole) 800 mg twice daily. After 1

month, both the pain and swelling were reduced. Further improvement was achieved after continued therapy and with 3 months follow-up.

Discussion

Mycetoma is caused by fungi or fungus-like bacteria. Two main groups of mycetoma have been identified; actinomycotic mycetoma and eumycotic or maduromycotic mycetoma depending on the causal agent. The causative organisms vary with geographical distribution, as does the site of involvement. Eumycotic mycetoma most commonly involves the lower extremities and is seen in rural areas amongst agricultural labourers working in primitive conditions because trauma especially following repeated contact with infected plants heralds the onset of disease [10,11]. Actinomycosis, on the other hand, affects the cervicofacial, thoracic and abdominal regions.

Mycetoma occurs in males 5 times more frequently than females [12], and of the last 5 reported cases from the Islamic Republic of Iran, 4 were males; our 2 new cases were both males. Poor personal hygiene, malnutrition, any history of trauma, wounds on bare feet and multiple systemic infections may predispose to the development of mycetoma [13]. The disease is commonest in the third and fourth decades of life, and it occurs mainly among farmers, herdsmen, field workers and those who walk bare-foot.

A classic case of mycetoma usually presents with a chronic indurated ulcerated plaque associated with swelling on legs, with discharge that contains yellowish granules [14]. The differential diagnosis of mycetoma includes botryomycosis, chromoblastomycosis, other deep fungal and tuberculosis infections, syphilis, yaws, elephantiasis and malignancy [1].

Radiographic examinations can reveal bone involvement, but basic radiological changes of osseous structures include osteoporosis, loss of cortical margin, erosion, lytic lesions and bony expansion [1,9]. Recently ultrasonography has been used in the differential diagnosis between mycetoma and other infections [12]. Differentiating eumycetoma and actinomycetoma is possible with focus on the colour of the granules. Microscopic evaluation of the pus discharge containing granules and pathology results of excision biopsy are required to identify the infective organism. But it should also be confirmed by culture [9,12]. Fine-needle aspiration is an effective method for rapid diagnosis of mycetoma, but it is valuable only in the case of aspiration of granules. Histopathological identification of the causative organism in the absence of positive culture has been shown to be useful in the diagnosis of mycetoma, but sufficient tissue is needed requiring deep surgery, which can result in spreading the disease [12,15].

The approach to therapy and prognosis of the disease depends on the infecting agent [16]. Patients in the early stage of mycetoma can be successfully treated medically without recourse to surgery [17]. Response to treatment is assessed by grades [18]. Grade 1: cured, all sinuses healed and swelling resolved; grade 2: greatly im-

proved, all sinuses healed and less swelling; grade 3: improved, most sinuses healed; and grade 4: no change or deterioration. The treatment most widely used is sulfonamides alone or in combination with other drugs for 6–18 months.

The 2 reported cases in our study were considered cured although the first patient had been affected for 33 years. Treatment remains a problem in chronic cases of mycetoma. In a study on 28 patients, only 9 were cured with medical treatment and 2 had relative improvement [6]. In another study, 4 cases out of 20 treated with streptomycin were completely cured. It has been emphasized that a quick response to medical treatment should not be expected and long-term treatment may be required [19]. Both cases in our study had a history of therapeutic and surgical treatment, which indicates the difficulty in diagnosing and curing the condition. However, correct diagnosis and appropriate treatment in our 2 cases highlight the fact that although eumycotic mycetoma needs prolonged and intensive antibacterial chemotherapy, even cases with extensive disease can be successfully treated.

Physicians should be aware that slowly spreading destruction of fascia, bones and tissues can be the key for diagnosis of mycetoma in the early stages.

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