

Mycetoma in the Western Region of Saudi Arabia

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ABSTRACT. Thirty-six (36) patients with histologically proven mycetoma were studied in three teaching hospitals in Jeddah between October 1984 and July 1990. The distribution of mycetoma in Saudi Arabia was also reviewed.

Keywords: Mycetoma, Madura foot, Saudi Arabia.

Introduction

Mycetoma is a condition with a worldwide distribution. It is endemic in tropical and subtropical locations such as parts of Africa, India, and the Middle East. Some recent reports^[1,2,3,4] show that mycetoma is not uncommon in Saudi Arabia. These reports, however, are from the eastern and central parts of the Kingdom alone. As far as we know, no report has been published from western Saudi Arabia. This report documents the occurrence of mycetoma in Jeddah, a coastal Saudi city on the Red Sea in the western region. The known overall geographic distribution and microbiological characteristics of mycetoma as seen in Saudi Arabia will also be reviewed.

Patients and Methods

A retrospective study was done on 36 patients who presented to three teaching hos-

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pitals in Jeddah between October 1984 and July 1990. The clinical and histological diagnosis of mycetoma was already confirmed. The hospital records were reviewed for age, sex, nationality, occupation, and socio-economic status. Clinical features, relevant laboratory findings, treatment modalities, and results of treatment were recorded and analyzed.

Published data on mycetoma from other centres in Saudi Arabia were also reviewed and analyzed along with the present series to determine geographic distribution, social factors, and microbiological pattern. The paraffin blocks and/or surgical specimens were retrieved and subjected for further studies. Haematoxylin and eosin (H&E), silver nitrate, and Periodic-Acid-Schiff (PAS) stains were used in preparations.

Results

Age and Sex Distribution: There were 30 males and six females, (a male:female ratio of 5:1). The age ranged from 19 to 55 years with a mean of 34.4 years.

Nationality: Most patients (80.5%) were non-Saudi. Of these, 24 (66.6%) were Yemeni living in the western region of Saudi Arabia, two (5.5%) were Sudanese, two (5.5%) were Somalis, and one (2.8%) was from Chad. Only seven (19.4%) patients were Saudi.

Occupation and Social Status: Twenty-six (26) (72.2%) were farmers, two (5.5%) were drivers, and two (5.5%) were soldiers. The six (16.6%) females were housewives (Table 1). All the patients were of low socio-economic status.

Clinical Presentation: The duration of symptoms ranged from four months to 30 years. All 36 patients presented with a swelling in the affected anatomical location. Twenty-four (24) (66.6%) patients gave a history of multiple discharging sinuses. There was an associated history of trauma in six patients (16.6%). Four patients (11.1%) had ulcers in the affected part of the body (Table 2).

TABLE 1. Occupations of the patients.

Occupation	No. of Patients	Percentage
Farmers	26	72.2
Drivers	2	5.5
Soldiers	2	5.5
Housewives	6	16.6

TABLE 2. Clinical presentation.

Presentations	No. of Patients	Percentage
Swelling	36	100.0
Sinuses	24	66.6
Trauma	6	16.6
Ulcers	4	11.1

The lower limbs were affected most: feet in 31 patients (86.1%), knees in two patients (5.5%), and legs in one patient (2.8%). Other locations were chest wall and hand, one in each (2.8%) (Table 3). Three patients (10%) had, in addition, enlarged inguinal lymph nodes. These were patients with eumycetoma infection.

TABLE 3. Location of lesions.

Site	No. of Patients	Percentage
Lower limb	34	94.4
Foot	31	86.1
Knee	2	5.5
Leg	1	2.8
Chest wall	1	2.8
Hand	1	2.8

Investigations: Radiological examination revealed bony destruction in six (16.6%) patients whose feet were diseased, while two (5.5%) had chronic osteomyelitic changes. One patient (2.8%) had a knee joint effusion. However, in the majority of patients, 27 (75%) had no bone involvement (Table 4). No haematological abnormalities were encountered in any of these patients.

TABLE 4. Radiological findings.

X-Ray	No. of Patients	Percentage
Normal	27	75.0
Lytic lesions	6	16.6
Osteomyelitis	2	5.5
Knee effusion	1	2.8

Histological Diagnosis: Sections from the paraffin blocks of the patients were stained with H&E, silver nitrate, and PAS. They were studied by an independent histopathologist. The findings were consistent with the original diagnosis, i.e., 30 (83.3%) cases of eumycetoma and six (16.6%) cases of actinomycetoma.

Treatment: The treatment modalities utilized included local excision alone, local excision with chemotherapeutic agents, or amputation. Sixteen (16) patients (44.4%) had local excision with use of multiple chemotherapeutic courses. Fifteen (15) (41.6%)

had local excision alone. Three had below-knee amputations while two had forefoot amputations (13.9%) (Table 5).

TABLE 5. Treatment modalities.

Therapy	No. of Patients	Percentage
Local excision and chemotherapy	16	44.4
Local excision	15	41.6
Amputations	5	13.9

The chemotherapeutic agents used included trimethoprim-sulphamethoxazole, penicillin, ketoconazole, dapsone, streptomycin, tetracycline, and griseofulvin used singly or in different combinations.

Results of Treatment: There was no mortality. However, 19 (52.7%) patients had recurrences during follow-up which ranged from one month to 41 months. There were no recurrences in patients treated by amputation.

Distribution of Mycetoma in Saudi Arabia: An analysis of all published data on mycetoma in Saudi Arabia up to 1990 revealed a total of 89 cases. The patients were from various parts of the central and eastern regions of Saudi Arabia. No similar reports from the western region. Adding 36 cases of the present series makes a total of 125 patients. Forty-five (45) patients (36.0%) were from the central region, 45 (35.2%) were from the eastern region, and 36 (28.8%) were from the western province accordingly (Table 6).

TABLE 6. Mycetoma in Saudi Arabia.

Author	Province	No. of Patients
Butler <i>et al</i> [3]	Central	9
Kubba & Satir [4]	Eastern	10
Aramco [1]	Eastern	11
Mahgoub & Gumma [13]	Central	5
Bendl <i>et al</i> [2]	Central	31
Vanugopal <i>et al</i> [9]	Eastern	23
Present study	Western	36
	TOTAL	125

Discussion

Mycetoma (Madura foot or maduromycosis) is a chronic fungal or bacterial infection which affects the skin, subcutaneous tissues, and bones. It was first described in the region of Madura, India in 1862 by Gill^[5]. Mycetoma has a worldwide distribution with endemic in tropical and subtropical countries in Africa, Asia, Central America, South America, and the Middle East^[6,7]. Mycetoma in of two types: eumycetoma, a true fungal infection, and actinomycetoma caused by bacteria.

The earliest reports of mycetoma in Saudi Arabia were from the eastern region where three cases were encountered over a 14-year period^[1]. Between 1984 and 1990, 89 cases were reported from various parts of eastern and central Saudi Arabia. The present paper reports the occurrence of mycetoma in Jeddah, a cosmopolitan coastal city with a large immigrant population from Yemen and the African countries. Located on the eastern shore of the Red Sea, Jeddah has an arid climate, suitable for mycetoma to be endemic. The clinical features of our patients are similar to those reported from Sudan^[6], South India^[8], and Saudi Arabia^[1,2,3,4,9]. Eighty percent (83.3%) were males and 26 (72.2%) were farmers. The six women in this series (16.6%) were spouses or relatives of farmers exposed to similar rural environmental factors. It is of interest to note that 29 (80.5%) of all patients in this series were non-Saudi. Twenty-four (24) (66.6%) were Yemenis and five (13.8%) were Africans. All patients in this series were of low socio-economic status.

The foot as a common site of infection (86.1%) is in accordance with the known mode of infection of mycetoma^[6]. Other locations were by no means exempt. Once infection has established, it invades the skin and subcutaneous tissues and multiple discharging sinuses become manifest as it was in 66.6% of our cases. The fungus grains are variable in size, shape, and texture. In the present series 18 patients had black, three had yellow, and one had mixed grains. Associated trauma was found in six (16.6%) of the patients but bone destruction and osteolytic changes were less encountered.

Deficient cell-mediated immunity was demonstrated by Mahgoub *et al*^[10]. However, Bendl *et al*^[2] did not show an obvious deficiency in cell-mediated or humoral immunity.

El-Hassan and Mahgoub^[11] drew attention to the regional lymph node involvement that sometimes accompanies mycetoma. They suggested that this was "almost certainly contributed to stimuli by fungal antigens". All three patients who had regional lymphadenopathy in this series were with histologically proven Eumycetoma. The fungi reached the inguinal lymph nodes in only one case.

For many decades, the treatment of mycetoma rested heavily on amputations of the extremity involved. Even so, recurrences were known to occur locally or in the regional lymph nodes. More recently, the efficacy of certain chemotherapeutic agents in the treatment of various forms of mycetoma have been documented^[12,13].

In 1976 Mahgoub^[12] achieved a 63.2% cure rate in 144 actino-mycetoma patients treated with various chemotherapeutic agents. Twenty-one percent showed great improvement. Treatment was successful even when there was advanced bone involvement. The most successful drug combination was dapsone and streptomycin or sulfamethoxazole-trimethoprim (cotrimoxazole) and streptomycin. Only two out of 20 patients (10%) with Madura mycetoma responded to this drug combination. Surgical treatment alone was not superior to surgery combined with griseofulvin and penicillin.

About 20% of these patients developed recurrences within 60 months of follow-up. More recently, ketoconazole has been found to be of great benefit in eumycetoma with a 38% cure rate^[13].

Mycetoma caused by *Madurella madurae* is the commonest variety found in this series, Jeddah, and other parts of Saudi Arabia, as confirmed in this report. This is keeping with the known geographical and ecological preference of the organism for arid zones with low rainfall levels, sandy soils, and thorny vegetation. Understandably, *Nocardia* species and *Madurella grisea* are not featured in this series as they are commonly encountered in tropical rainforest belts and in Latin America^[9]. Bendl^[2] and Venugopal^[9] reported one case of *Nocardia asteroides* and two cases of *Madurella grisea* in Saudi Arabia, respectively.

Only one case of *Pseudallescheria boydii*, commonly encountered in North America and Europe, was encountered in the entire Saudi experience^[1].

In early stages before development of characteristic discharging sinuses, the diagnosis may be different, especially if it occurs in an unusual site. It should be considered in the differential diagnosis of all painless subcutaneous swellings, especially in the tropics^[9].

Mycetoma is a particularly devastating condition, frequently requiring prolonged chemotherapeutic measures and often ending in amputation of the affected part or other surgical maneuvers. Considering the known epidemiological pattern of the condition, a further improvement in public health measures, particularly in the rural areas, will be of great help in eradicating this condition in endemic areas.

In the present series, surgical excision was the main form of treatment, used either alone (41.6%) or in combination with chemotherapeutic agents (44.4%). The 52.7% recurrence rate in this series illustrates the high morbidity associated with mycetoma.

In 1984, Mahgoub^[13] stated that there is no longer any justification for mutilating surgery or amputation in any type of mycetoma before giving medical treatment a genuine trial. The only problem with ketoconazole is its high price which, in many poor countries, cannot be afforded. The treatment should be continued for 12 months or even longer if the bone is involved. Recurrence is likely with irregular use of drugs. Patients must be closely followed up for development of hepatotoxicity and until they become serologically negative by the immuno-diffusion and counter immuno-electrophoresis tests.

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الورم الفطري (Mycetoma) في المنطقة الغربية، المملكة العربية السعودية

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المستخلص. تم دراسة ستة وثلاثين حالة من الورم الفطري Mycetoma جمعت من ثلاثة مستشفيات تعليمية في جدة بين أكتوبر ١٩٨٤م ويوليو ١٩٩٠ ومعظم الحالات (٥ . ٨٠٪) من غير السعوديين، وأكثرهم (٢ . ٧٢٪) من المزارعين، وقد تبين أن العامل الفطري كان من الفطور الحقيقية -Eu mycetoma في ٢ . ٨٣٪ من الحالات. هذه في تقديرنا الدراسة الأولى في المنطقة الغربية في المملكة العربية السعودية، وقد تضمنت توزيع الورم الفطري في المملكة العربية السعودية.