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A CASE REPORT OF MADUROMYCOSIS (*PSEUDALLESCHERIA BOYDII*) INFECTION TREATED WITH KETOCONAZOLE

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ABSTRACT

A case of mycetoma of the arm caused by *Pseudallescheria boydii* in a 56-year-old woman is described. Response to oral administration of ketoconazole was successful.

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INTRODUCTION

Mycetoma is a chronic granulomatous disease characterized by localized swelling, multiple discharging sinuses and presence of granules in the exudate or the tissues.[1]

Usually the condition begins as a painless nodule at the site of previous trauma. This primary nodule softens and discharges pus.[2] In chronic cases the sinuses may extend deeply into the muscles and bones.[2] The incubation period varies from one week to several months.[2] The disease results from the traumatic implantation of the organisms from the soil, actinomycetes (higher bacteria) or eumycetes (true fungi) into the tissues.[3,4] One of the eumycetes is *Pseudallescheria boydii* that is usually found in soil and is an opportunistic pathogen.[5,6] Occasionally this fungus can cause pulmonary and central nervous system involvement and disseminated infections, particularly in patients with a possible predisposing underlying condition.[7] The treatment of the cases depends on the causative agents and organ involvement such as bones and central nervous system.[8] Infections with the eumycetes are generally resistant to medical therapy.

Among this group *Pseudallescheria boydii* has proven sensitive to miconazole and a number of other, lesser well known antifungal agents *in vitro*.[4] Ketoconazole which is a new antifungal agent, belongs to the imidazole family which together with clotrimazole, econazole, and miconazole, has demonstrated the same therapeutic effects.[8] Koren (1983), Mujica, et. al. and Galgini, et.al. (1984) treated cases of mycetoma

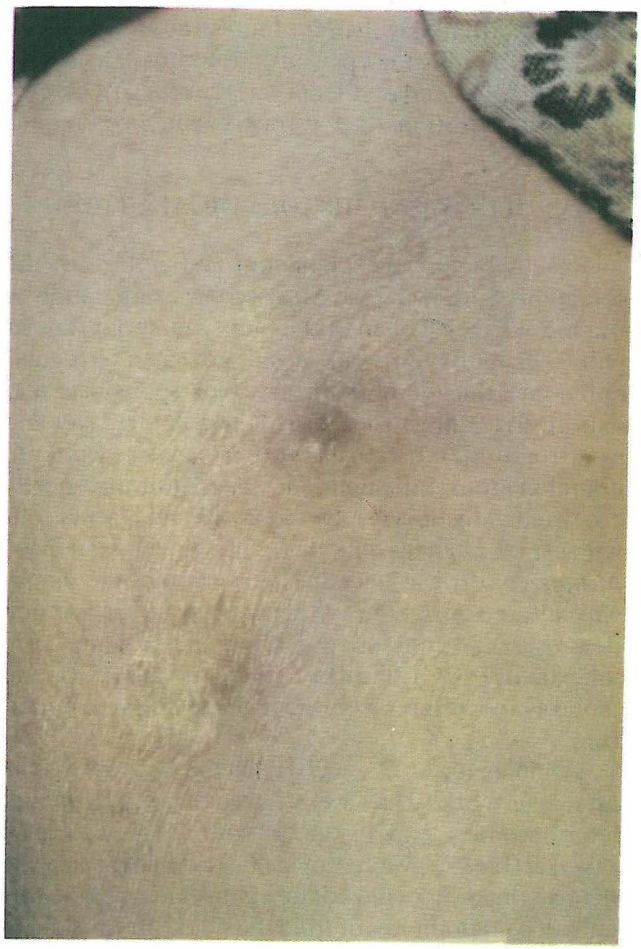

Fig.1. Maduromycosis caused by *Pseudallescheria boydii*.

caused by *P.boydii* with ketoconazole.[9,10,11] In the present study, a case of mycetoma, in which *P.boydii* was the causal agent, has been reported and successfully treated with oral ketoconazole.

CASE REPORT

A 56-year-old woman from Tehran was admitted to the Department of Dermatology, Razi Hospital, Tehran University of Medical Sciences in April 1987, with a pruritic,-erythematous, edematous lesion with secretion of a serosanguinous exudate on her left arm (Fig. 1). She stated that she had been on a country picnic 18 months before she developed a papule with pruritus, irritation and swelling on the left arm. It gradually became a small nodule that softened with discharge of pus and blood. No bone destruction was found in roentgenograms.

The patient was referred to the Medical Mycology Department, School of PublicHealth, Tehran University of Medical Sciences for further investigation. Histological section of the biopsy material showed a mixed granulomatous reaction and the culture of the tissue revealed growth of *Pseudallescheria boydii*. The patient was treated with oral administration of ketoconazole 200 mg/day for a period of 6 months. The lesion became limited and erythema was reduced after treatment but secretion of a serous exudate continued.

MYCOLOGY AND HISTOLOGICAL STUDIES

Results of direct examination and culture of the secretions were negative. Microscopic examination of the histological section of the biopsy material stained with heamatoxylin and eosin revealed irregular epidermal thickening with areas of spongiosis and exocytosis leading to absence of the granular layer and a parakeratotic crust. In the corium there was scattered granulomatous inflammation. The infiltrate mostly consisted of histiocytes, lymphocytes and plasma cells with a few Langhans type giant cells. Acid fast stain for *M.tuberculosis* was negative. A portion of the biopsy was cultured on Sabouraud dextrose agar,[5] Sabouraud dextrose agar containing chloramphenicol and cycloheximide (SCC), brain heart infusion agar (BHI) and blood agar. Cultures were incubated at room temperature.

Colonies on SCC and BHI media became visible after one week. They were floccose, initially white, later becoming gray in colour, with a dark brownish reverse (Fig.2). Microscopically, branching septate hyphae, bearing ovoid conidia with thick walls at the ends of conidiophores or from the side of the mycelium were seen (Fig. 3)

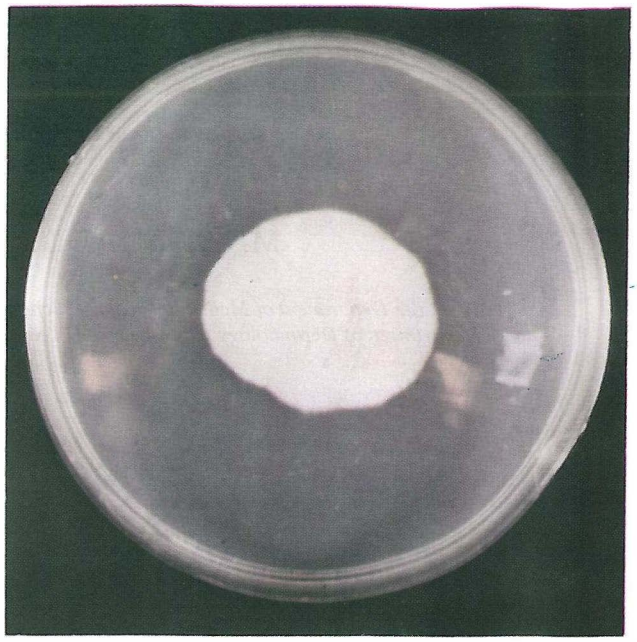

Fig.2. Colony of *Pseudallescheria boydii* on a Sabouraud dextrose agar containing chloramphenicol and cycloheximide disc, 1 week old at room temperature.

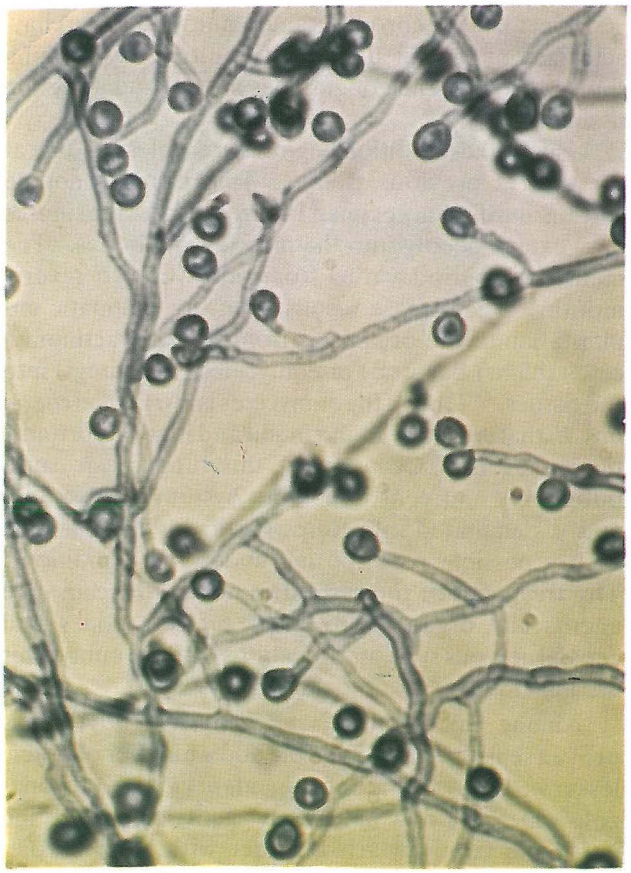

Fig.3. Branching septate hyphae, bearing ovoid conidia with thick walls at the ends of conidiophores or from the sides of the mycelium of *Pseudallescheria boydii*, lactophenol cotton blue mount.

DISCUSSION

The present case of mycetoma due to *Pseudallescheria boydii* is the third case reported in Iran. The first two cases were diagnosed in the Medical Mycology Department, School of Public Health of Tehran University of Medical Sciences during 21 years from 1961 to 1982.[12]

Although *Pseudallescheria boydii* is generally an opportunistic pathogen most commonly seen in patients demonstrating the triad of trauma, antimicrobial therapy, and corticosteroids,[13] our patient did not have a history of trauma in her country picnic. The discharge did not contain granules, but in histologic examination of the biopsy specimen a granulomatous reaction was seen, and culture of the tissue showed *Pseudallescheria boydii*.

Clinical reports suggest that amphotericin B is ineffective in the treatment of the patients. *P. boydii* usually demonstrates marked *in vitro* resistance to both amphotericin B and flucytosine. Ketoconazole, the new imidazole-derivative anti-fungal agent has been used for treatment of *P. boydii* infection in several cases.[9,10,11] In the present case, after treatment with ketoconazole, the lesion became limited and erythema was reduced but secretion of a serous exudate continued.

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