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CASE REPORT

Multiple Subcutaneous Mycetomas caused by *Pseudallescheria boydii*: Response to therapy with oral potassium iodide solution

Fida A. Khan [a,b,*], **Shahrukh Hashmi** [c], **Arif R. Sarwari** [b]

[a] *Ohio Valley Medical Center, Section of Infectious Diseases, 2000 Eoff Street, East Building, 4th Floor, Wheeling, WV 26003, United States*
[b] *West Virginia University, Section of Infectious Diseases, Morgantown, WV 26505, United States*
[c] *Aga Khan University Department of Medicine, Stadium Road, Karachi, Pakistan*

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Summary We describe the case of a sixteen-year-old male who presented with multiple subcutaneous mycetomas proven on culture to be secondary to *Pseudallescheria boydi.*, The lesions responded completely to oral potassium iodide solution. To our knowledge this has never been reported in humans.

Case report

A 16-year-old previously healthy boy presented with multiple subcutaneous masses on his neck, back, chest and abdomen. Eight years ago he had developed a small, non-tender, nodular mass on the left temporal aspect of his head with no preceding history of trauma or injury. The mass gradually increased in size to 2–3 cm in diameter and subsequently developed a sinus tract. The lesion later healed in two months but the patient was left with extreme scarring. About one and a half year later a similar mass appeared in the same location. Within two months, further subcutaneous lesions also developed over his neck, back, chest and abdomen; most resolving with formation of sinus tracts and keloid formation. No associated fever or systemic symptoms were reported. Three years before presentation, he had been prescribed itraconazole, which he took intermittently for six months with resolution of nodules, only to have them reappear two months after completion of therapy.

On examination, he appeared comfortable, with stable vital signs. Numerous subcutaneous masses involving the neck, trunk, arms and thighs ranging from 2 to 15 cm were noted (Fig. 1). These lesions were soft, fluctuant, with no warmth, or erythema. A few had sinus tracts with purulent drainage. The remainder of the physical exam was normal with no hepato-splenomegaly.

Initial laboratory data showed hemoglobin of 10.9 g/dL, white blood cell count of 13.6 K/UL and platelet count of 295,000 K/UL Peripheral blood smear was significant for anisocytosis, hypochromia, and microcytosis with mild eosinophilia. A bone marrow aspirate was normal. Lesions of

* Corresponding author. Tel.: +1 304 293 3306; fax: +1 304 293 8677.
E-mail address: fkhan@ovrh.org (F.A. Khan).

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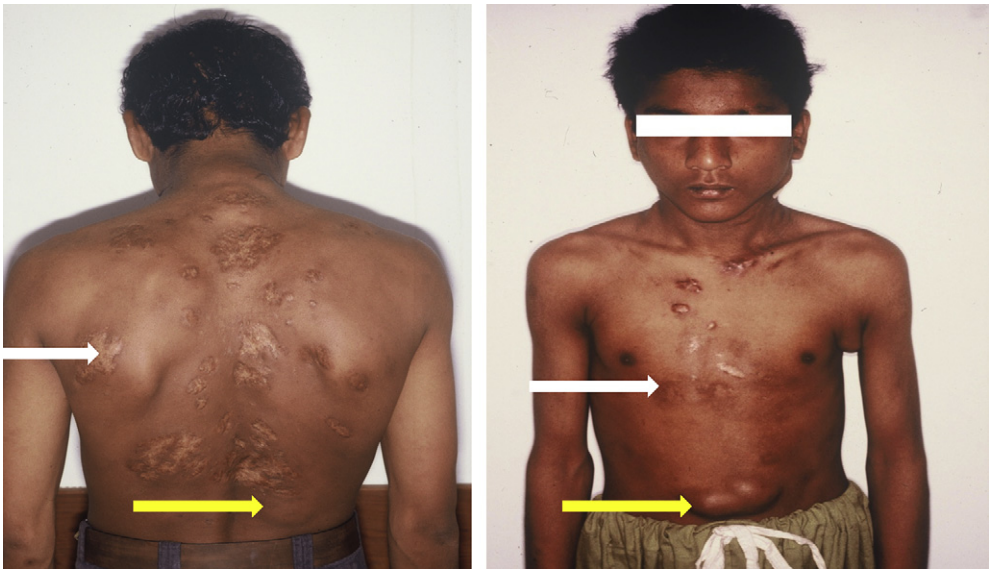

Fig. 1 Subcutaneous; Mycetomas before treatment Keloid formation prior lesions (white arrows) and Mycetoma (yellow arrows).

neck and abdominal wall were aspirated (Fig. 2), and both specimens grew out *P. boydii*, and the presence of fungus in the aspirate was confirmed using histologic stains (Fig. 3). A diagnosis of multiple subcutaneous mycetomas due to *P. boydii* was made and the patient started on therapy with oral itraconazole with periodic needle aspirations. He was, however, unable to afford the prescribed therapy for more than a month and was switched over to standard doses of Potassium Iodide saturated solution based on the Sporotrichosis literature i.e. 5 drops(1 ml) three times a day and subsequently increased to 30 drops each dose. Patient was followed about every 3 months for over a period of over two years with complete resolution of lesions (Fig. 4).

Discussion

Carter introduced the term Mycetoma, which refers to a clinical entity, first described in ancient Sanskrit literature, in 1860. Clinically this is characterized by localized, chronic, progressively destructive swelling, which involves the cutaneous and subcutaneous tissues, occasionally extending to fascia and bone.[1,2]

Fig. 2 Aspiration of purulence from Neck Lesion.

Lesions are composed of suppurating abscesses, granulomata and draining sinuses with ''grains'', nature of which depends on the etiology. Mycetoma can be caused by soil-inhabiting bacteria (actinomycetoma) or fungi (eumycetoma). *P. boydii* belongs to the genus Scedosporium and is found in soil worldwide. It grows cotton-like colonies with septate hyphae and forms white grains in mycetoma. It can enter the host by an airborne route causing respiratory disease (Pseudallescheriasis) or by a direct inoculation in a penetrating injury causing Mycetoma. It is a common causative agent of mycetoma in the United States. Extracutaneous sites are only rarely involved. *P. boydii* has been implicated as a causative agent in infections of the lung, sinuses, joints, glands (parotitis, thyroid abscess, prostatitis) and endocarditis.[1,3,4]

Disseminated disease usually occurs in immunocompromised patients.[2,5] In Mycetoma *P. boydii* enters tissue after a local penetrating trauma. It progressively involves and destroys connective tissue and bone which results in the formation of a suppurative granuloma, made up of inflammatory cells along with distinctive grains embedded in the abscess.

Initially, the lesion is a small, painless subcutaneous nodule that progressively enlarges, becomes phlegmonous, and finally ruptures to the surface with sinus tract formation.[1,2] The discharge is usually serous, but may be serosangious or purulent. As infection progresses, similar lesions appear on adjacent parts. Infection follows a chronic course, with damage to the underlying tissues. Super infection with bacteria can occur and may be life threatening.[6] *P. boydii* is apparently unable to produce an infection in an immune competent host except after a barrier break or traumatic implantation.[7]

The medical management of mycetoma, especially eumycetoma, has not been very successful. Survival rates among patients with disseminated or CNS disease have been