Pseudomycetoma Due to *Trichophyton schoenleinii* Occurring in Two Brothers

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A 14-year-old boy and his 16-year-old elder brother, who had generalized tinea corporis for 1 and 3 years respectively, developed multiple discrete, non-tender soft tumors on the scalp, forehead, neck and extremities. The boys were well nourished and had no systemic diseases. Histopathologic examination of the subcutaneous nodules revealed a well encapsulated granuloma containing lobulated granules characteristic of mycetoma; these granules consisted of septated fungal hyphae with vesicles. The dermatophyte isolated from the tumors was identified as *Trichophyton schoenleinii*. (Ann Dermatol 1:102-106, 1989)

Key·Words: Pseudomycetoma, *Trichophyton schoenleinii*

Dermatophytes have been demonstrated infrequently in viable tissues such as the dermis, subcutis, lymph nodes, bones and other organs.[1-6] Very rarely, mycetoma-like lesions caused by dermatophytes, known as pseudomycetoma, have been reported.[7-13]

To our knowledge, mycetoma-like nodular masses have not been shown to be caused by *Trichophyton (T.) schoenleinii*. We present two cases of two brothers who had pseudomycetoma on the scalp, forehead, neck and extremities caused by *T. schoenleinii*.

REPORT OF CASES

Case 1: A 14-year-old Korean boy presented with indolent masses on his scalp, forehead and neck and fistula-forming masses on his wrist present for 4 years. His illness began at the age of 8 years with oozing and crusted dermatosis on his scalp. This dermatosis spread over the forehead, neck, extremities and trunk and it cleared 1 year after the onset of the above masses. Multiple indolent nodules were present on his scalp and wrists from the age of 10 years, and the number and size of the nodules increased progressively. When the masses ruptured, they drained serosanguineous matter. There was no episodes of complete remission. His elder brother (Case 2) had the same type of skin lesions.

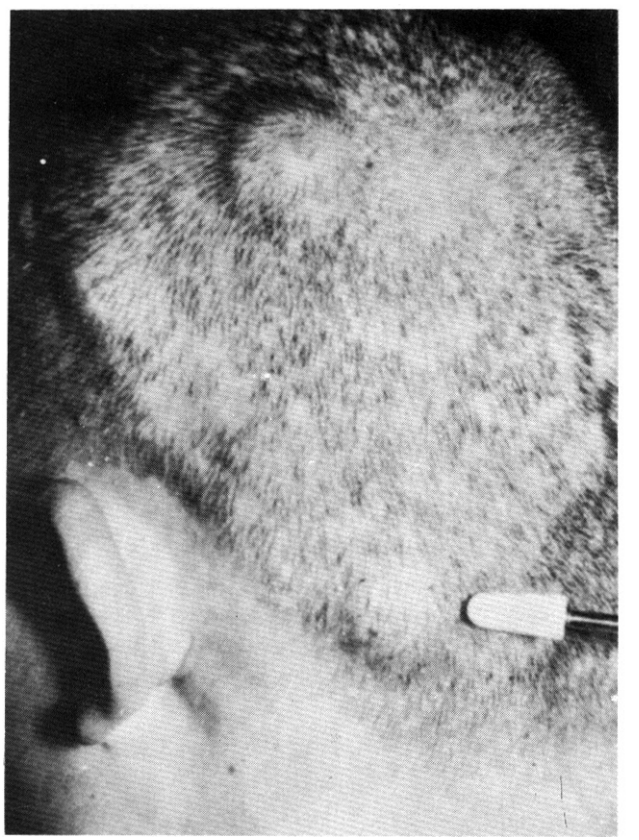

Fig. 1. Nodules on the scalp of Patient 1 resembling lipomata or cysts.

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On physical examination, multiple discrete, nontender, movable masses were seen on his scalp, forehead, neck and extremities. They varied from 1 cm to 7 cm in diameter and they resembled lipomata or cysts (Fig. 1). An examination of the regional lymph nodes showed no enlarged nodes. All finger nails on his right hand and the great toe nail on his left foot were greyish white with subungual hyperkeratosis. The remainder of the physical examination showed no further abnormality.

Findings from the laboratory studies of the patient were negative or normal. These included routine examinations of stool and urine, a liver function test, VDRL, and fasting blood sugar. Serum chemistry values, chest roentgenogram and serum immunoelectrophoresis were normal. Anti-streptolysin O titer was 1:340 and 17-ketosteroid was 10.7mg/24h (normal, 4-9 mg/24h in the 12 to 16 age group). Complete blood cell count, except for a mild eosinophilia of 5%, was within the normal limit. Skin tests with DNCB, PPD, and trichophytin were positive. Comparisons of mean percentages of total and active T-cell populations revealed no significant differences between this patient and the normal control groups.

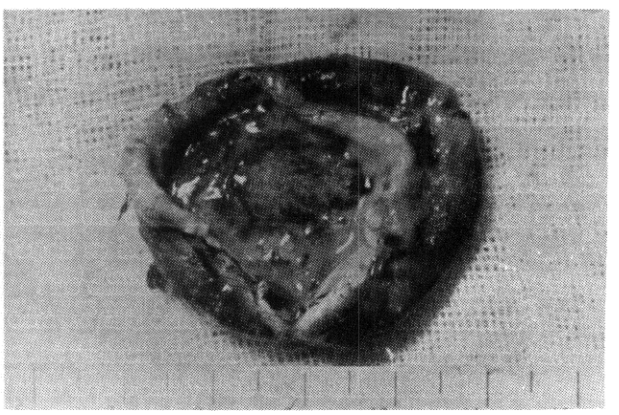

Fig. 2. The excised cystic mass from the neck revealed well encapsulation.

The excised solid mass from the scalp was well encapsulated, as was the excised cystic mass from the neck (Fig. 2). Vessels were abundant in the cystic wall and within the cystic space there was serosanguineous fluid containing yellow granules. Microscopic sections showed numerous fungal grains of approximately 70-180μm in diameter, consisting of broad, septate hyphae.

Microscopic examination of a specimen from the solid mass revealed a large well encapsulated granuloma in the dermis. The large, faintly eosinophilic stained, lobulated granules resembling grains of mycetoma were found in the areas of caseous necrosis. The granules were surrounded by granulomatous inflammation, including macrophages and foreign body giant cells (Fig. 3,A). On staining with periodic acid-Schiff stain, the granules showed septate hyphae with branches and frequent vesicles (Fig. 3,B).

Specimens from the nodules were cultured on Sabouraud dextrose agar and incubated at 25°C for three weeks. The isolate grew very slowly and the colonies were yellowish-white and irregularly heaped-up and folded and the colony reverse was colourless (Fig. 4). Microscopic observation revealed very coarse,

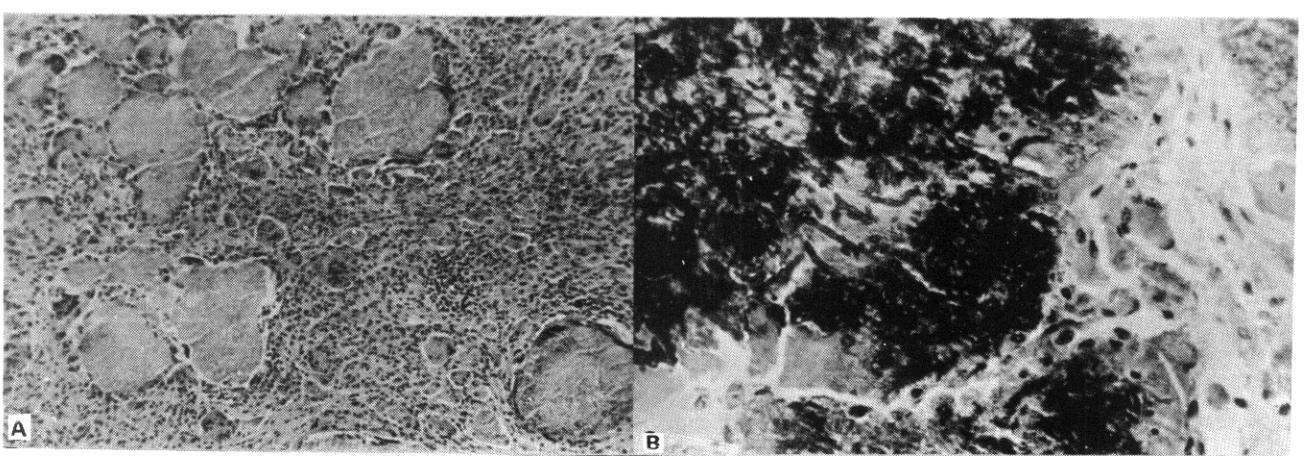

Fig. 3. A, histopathology of the nodule from Patient 1, demonstrating faintly stained lobulated granular masses in a large well encapsulated granuloma in the dermis. The granules are surrounded by granulomatous reaction containing many marcrophages and foreign body giant cells (hematoxylin-eosin stain, ×100). B, higher magnification demonstrating accumulation of numerous nterlaced fungal hyphae (periodic acid-Schiff reaction, ×400).

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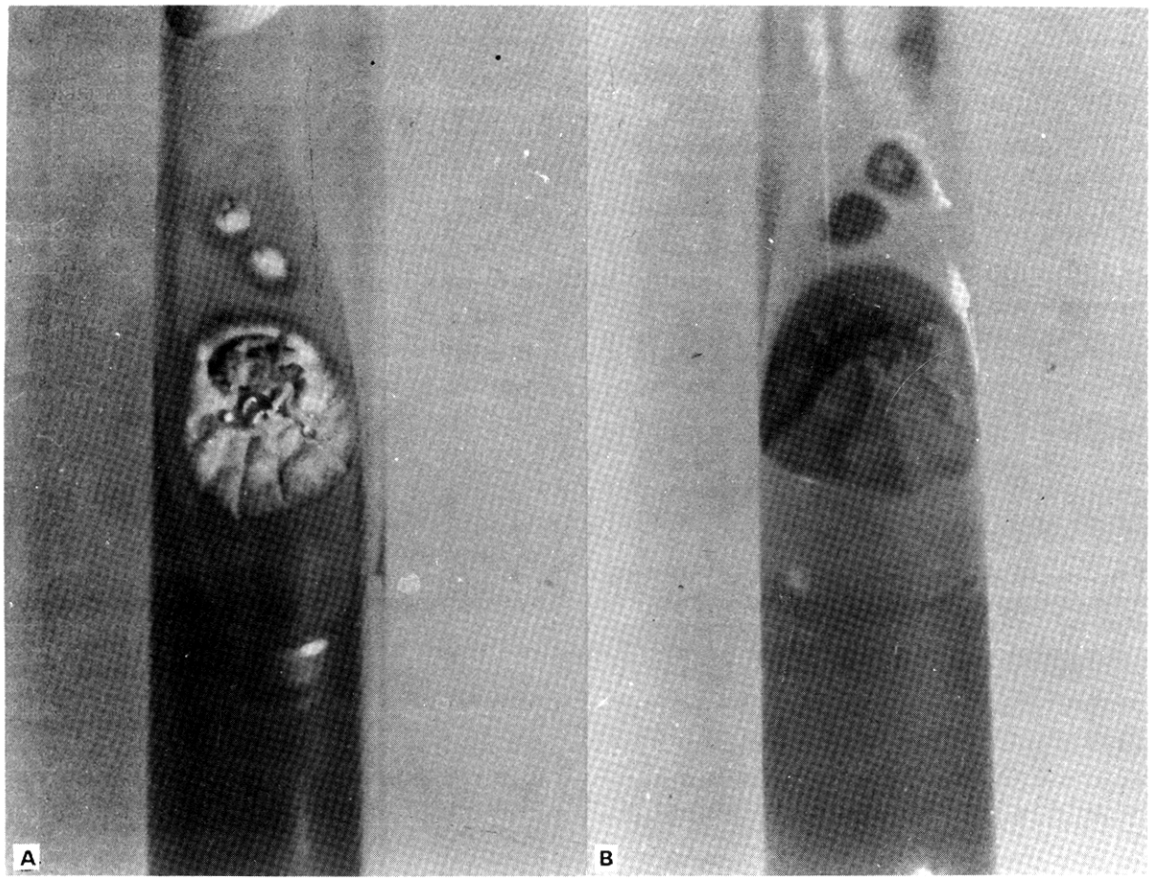

Fig. 4. A, slowly growing, yellowish-white and irregularly heaped-up and folded colonies appeared after 3 weeks of cultivation on Sabouraud's dextrose agar at 25°C. B, reverse was colorless.

ramified hyphae and partly chandelier-like terminal hyphal branches were present. The fungus was tentatively identified as *T. schoenleinii*. The isolates were sent to Dr. Michael Dorn of the University of Munich, Federal Republic of Germany and to Dr. Renoo Kotrajaras of the Institute of Dermatology, Bangkok, Thailand for identification. Both confirmed that the isolate from the scalp mass was more compatible with *T. schoenleinii*. than with any other species of dermatophyte.

Surgical excision and nail extraction were done and treatment was commenced with griseofulvin 500 mg per day.

Case 2: A 16 year-old Korean male presented with two painless masses on his scalp. Approximately 6 years ago, he experienced scaly patches on his scalp which spread over his entire body surface area and cleared 3 years later. Two years ago, one palpable mass developed in the occipital area and it enlarged progressively, thereafter other masses appeared.

On physical examination, non-tender, movable soft masses were noted on the scalp. They resembled lipomata and were 2.5×2.5 cm and 1×1 cm in size. All finger nails on both hands showed greyish white dystrophic changes with subungual hyperkeratosis.

Findings from laboratory studies of this patient were negative or normal. These included complete blood cell count, routine examinations of stool and urine, a liver function test, VDRL, fasting blood sugar, serum chemistry, anti-streptolysin O titer, and 17-ketosteroid. The chest roentgenogram and serum immunoelectrophoresis were also normal. Skin tests with DNCB, PPD, and trichophytin were positive. Comparisons of mean percentages of total and active T-cell populations revealed no significant differences between this patient and the normal control groups.

Two solid masses excised from the scalp revealed good encapsulation. Both nail and skin biopsies yielded *T. schoenleinii*. on the culture. Histopathologic findings were the same as in Case 1.

DISCUSSION

T. schoenleinii. produces a distinctive and remarkable type of tinea capitis called favus. The granulomatous lesions shown in these cases do not parallel the usual clinical manifestations of favus due to *T. schoenleinii.* The dermatophytes are generally fungi that are parasitic on keratin portions of the epidermis, hair, and nails; they do not ordinarily penetrate deeper than the basal cell layer.[14] They can, however, invade the deep corium sometimes through an infection of the pilosebaceous apparatus. Since it was described by Majocchi 100 years ago, a few cases of deep granulomatous cutaneous infections due to dermatophytes such as *T. violaceum,*[1-3] *T. verrucosum,*[4] *T. rubrum*[5] and *T. tonsurans*[6] have been reported.

Mycetoma is a clinical syndrome caused by *Actinomycetes* and fungi that is characterized by suppurating abscesses, granulomata, and draining sinuses with the presence of "grains". Many cases of this disease had been observed in the province of Madura in India; hence, the name madura foot. The involved organisms may also cause other clinical diseases such as actinomycosis, mycotic granuloma, and phaeohyphomycosis, but only when the above criteria are met is the diagnosis of mycetoma valid. However, a clinical condition similar to mycetoma called botryomycosis may be caused by a number of other bacterial species and a condition called fungus balls may be caused by opportunistic mold pathogens such as species of *Aspergillus* and *Cladosponum*[14] Recently, similar processes caused by dermatophytes were reported, known as pseudomycetoma, this is rare.

Pseudomycetoma may be induced by a mechanism in which the hair follicles are ruptured due to extension of the growth of the dermatophytes, and the contents are released into the the surrounding dermis. Due to a hypersensitivity response to the dermatophytes, a granulomatous reaction occurs with deposition of amorphous material around the fungus. The morphology of fungal grains reported for pseudomycetomas due to different dermatophytes are identical.[12] The closely septated hyphae are typically imbedded in clear eosinophilic cement. The terminal or intercalary enlargement of hyphae, resembling vesicles, is frequent.[15] As the causative organisms of this distinctly unusual disease, *T. tonsurans,*[7,8] *T. violaceum,*[8] *T. verrucosum,*[9] *M. ferrugineum,*[10] *M. canis,*[11,12] and *M. audouinii*[13] have been reported. *T. schoenleinii* infection of skin is not uncommon, but the appearance of the mycetoma is hitherto unreported.

Dermatophytes infections in human and animals are significantly influenced by a disturbance of cell-mediated immunity. When the host's immunologic system is altered, dermatophytes may produce a more widespread and virulent disease in the affected hosts by lymphatic or hematogenous dissemination.[16] In our patients, the skin tests with DNCB, tuberculin and trichophytin showed positive reactions and T cell counts were normal. The patients were well nourished and had no noticeable systemic diseases. Roth et al.[17] have suggested from their studies that serum factors prevent invasion of living tissue by the dermatophytes and that the decreases in fungistatic activity of the patient's serum allowed dermatophytes to penetrate into living tissue. At the present time, we do not know whether humoral factors might be responsible for the formation of fungal granuloma in living tissue.

In these cases, it is very interesting that the same type of lesion developed in brothers. *T. schoenleinii* is classified as a anthropophilic dermatophyte, that is, it inhabits the skin surface of humans.[18] This infection may occur concurrently in more than one member of a family by direct human-to-human contact. However, the sequential occurrence of these unusual clinical features of *T. schoenleinii* infection in brothers suggests that these patients have a specific defect in the genetic ability to respond to the specific antigenic components of *T. schoenleinii,* although we do have not any supporting laboratory evidence.

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