A Giant Dermatophyte Abscess Caused by *Trichophyton rubrum* in an Immunocompromised Patient

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ABSTRACT

A 73-year-old male who had been receiving immunosuppressive drugs for 15 years developed a nodule on the left buttock region. The nodule slowly grew into a 15-cm fluctuant multilocular subcutaneous cyst. Serum beta-D-glucan levels were high, and the yellow purulent fluid obtained from the cyst was positive for *Trichophyton rubrum*. Granuloma formation in the cyst wall and large abscesses in the central cystic area were found, and septated hyphae were observed in both tissues. The cyst was surgically removed, and followed by itraconazole treatment. Notably, the clinical manifestations closely resembled those of a huge atheroma.

Key words: dermatophyte abscess, dermatophyte granuloma, immunocompromised patient, serum beta-D-glucan, *Trichophyton rubrum*

Introduction

Dermatophytes are keratinophilic fungi that usually infect the stratum corneum, hair, and nails. However, dermatophytes occasionally invade the dermis, subcutaneous tissues, and internal organs, resulting in a condition called deep dermatophytosis.

We herein report a case of a giant dermatophyte abscess of the buttock caused by *Trichophyton rubrum* in an immunocompromised patient.

Case

A 72-year-old Japanese male developed a nodule on the left buttock without any trauma, and the nodule gradually grew over 2 years. The patient’s medical history included interstitial pneumonia 15 years prior and osteonecrosis of the left femoral head 10 years earlier. The patient experienced worsening of interstitial pneumonia 2 years earlier, for which he received pulse-steroid therapy followed by combined treatment with oral prednisolone and cyclosporine. Physical examination showed a well-demarcated, elastic, soft, partially fluctuant, multilocular subcutaneous
cyst measuring 15 cm in diameter (Fig. 1). There was no redness, tenderness, fistula formation, or purulent discharge. Clinical manifestations were not apparent on the surface skin of the cyst, and a small amount of scales was obtained by scraping the skin with a scalpel. KOH preparation of the scales revealed septated hyphae.

Computed tomography examination showed a subcutaneous multilocular cyst. A large amount of yellow purulent fluid was obtained from the cyst by needle aspiration, and KOH preparation of the purulent fluid revealed a large number of septated hyphae (Fig. 2). Laboratory tests revealed a white blood cell count of 11,300/µl (normal range, 3,900–9,700/µl), blood urea nitrogen of 24.0 mg/dl (9–21 mg/dl), creatine of 1.15 mg/dl (0.6–1.2 mg/dl), and C-reactive protein of 1.6 mg/dl (<0.2 mg/dl). Moreover, a high level of serum beta-D-glucan (984 pg/ml; normal range, 0–20 pg/ml) was observed.

Pathological examination showed a multilocular cyst, with the cyst wall forming granulomas composed of giant cells, histiocytes, and lymphocytes, and the interior of the cyst forming abscesses composed of neutrophils, with many septated hyphae. High-resolution images of the cyst stained with periodic acid-Schiff showed many spore chains and septated hyphae being phagocytosed by multinuclear giant cells and histiocytes (Fig. 3). Fungal cultures of the scales and abscesses yielded slightly raised, white to cream, downy, with a brown reverse colony. (Sabouraud’s glucose agar for 3 weeks at 27°C).

Fungal culture of material from concomitant tinea
pedis and tinea unguium was unsuccessful. Urease tests were negative. The nucleotide sequences of the internal transcribed spacer 1 (ITS1) region in the ribosomal RNA gene of the clinical isolates had 100% (317/317 bp) homology to that of *T. rubrum* standard strain ATCC 28191 (GenBank accession no. KF278457).

After admission, the patient was initially treated with incision and drainage of the lesion. However, drainage was difficult to perform because the lesion was multilocular and had thick septum. Therefore, the cyst was completely removed surgically, and oral itraconazole 50 mg/day was administered for 25 weeks. Postoperative ulcers were treated with negative pressure wound therapy for 2 weeks, and 18 weeks later, a scar developed (Fig. 6). The serum beta-D-glucan level finally fell to 18.5 pg/ml. The patient had not experienced recurrence after 2.5 years of follow-up.

**Discussion**

Deep dermatophytosis, also known as "Majocchi’s granuloma," is very rare, with only 100 prior cases reported in literature. Classification of deep dermatophytosis has still not been established due to the diversity of clinical manifestations of this disease. In deep dermatophytosis, the deep lesions are usually accompanied by superficial dermatophytosis. Fukushiro categorized deep dermatophytosis into the following four clinical entities: (1) dermatophyte granuloma (i.e., Majocchi’s granuloma), subcategorized as (1a) the localized form in which subcutaneous nodule formation is accompanied by pre-existing superficial dermatophytosis at selected skin sites, and (1b) in which subcutaneous nodule formation is accompanied by diffuse superficial dermatophytosis that may disseminate to internal organs and can cause fatality (for both of these forms, granuloma formation, but not abscess, is seen in the nodules); (2) nodular granulomatous perifolliculitis of the legs, in which formation of multiple chronic nodules surrounding hair follicles is observed unilaterally in the lower leg, representing a variant form of the clinical entity indicated in 1a; (3) dermatophyte abscess, consisting mainly of abscesses containing dermatophytes in the dermis and/or subcutis; and (4) dermatophytic mycetoma, for which the clinical manifestations include formation of a sinus tract that discharges exudates containing dermatophytic granules.

Our patient presented with symptoms similar to those of atheroma; a large volume of purulent fluid was pathologically observed within the septum-forming granuloma. Kobayashi et al. and Matsuzaki et al. reported cases similar to ours, which were diagnosed as tinea profunda cysticum and deep pseudocystic dermatophytosis, respectively. The present case may be categorized as dermatophyte abscesses according to the classification of deep dermatophytosis proposed by Fukushiro, because the pathological features consisted mainly of abscesses.

Fukushiro reviewed 32 cases of dermatophyte abscesses between 1966 and 1998, and Inaoki et al. subsequently reviewed 14 cases between 1999...
and 2015, including those reported by Kobayashi et al. and Matsuzaki et al. Additionally, Kim et al. reported similar cases in 2016. A summary of the clinical characteristics of these 47 cases indicated that 38 cases (83%) occurred in immunocompromised patients who presented with multiple nodules; 54% had involvement of the groin, buttocks, and lower extremities, and the majority had pre-existing superficial dermatophytosis. The causative pathogen of dermatophyte abscesses was *T. rubrum* in 47 (81%) cases, and serum beta-D-glucan levels were elevated in six cases. Our patient showed a significantly high level of serum beta-D-glucan (984 pg/ml), which provided an important clue for diagnosis (the serum beta-D-glucan level decreased with clinical improvement).

In the present patient who had been receiving immunosuppressive therapy for interstitial pneumonia for 15 years, the cutaneous lesion was difficult to notice. However, a culture of the scales on the surface skin of the cyst yielded *T. rubrum*, which was similar to that from the abscess. The fungus present in the superficial tissue was likely to have invaded the subcutaneous tissue during immunosuppressive therapy, causing formation of a subcutaneous cyst.

According to Nishiyama et al., fungal strains isolated from dermatophyte granuloma show higher levels of protease activity, despite the lower growth rate, than those isolated from tinea superficialis. This observation suggested that the fungi isolated from dermatophyte granuloma can survive and grow in the dermal environment despite morphological and functional changes, causing formation of a subcutaneous cyst.

In summary, we herein reported a case of a giant dermatophyte abscess in an immunocompromised patient. Notably, in our patient, clinical manifestations, including symptoms similar to those of atheroma, complicated the diagnosis of fungal disease, and the presence of significantly high levels of serum beta-D-glucan served as a diagnostic tool. Thus, dermatomycosis should be considered in any examination of patients who are receiving immunosuppressive drugs.

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**Conflict of interest**

None declared.

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