A Case of Subcutaneous Infection with *Trichosporon cutaneum* in a Kidney Transplanted Patient

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INTRODUCTION

Tricosporon(*T.*) *cutaneum* is a member of the subfamily *Trichosporideae* in the family of *Cryptococcaceae*. It is now regarded as the same species as *Trichosporon beigelii*. It is widely distributed in soil, and is sometimes a part of the normal flora of the human skin, nail and mouth[1]. It also may rarely colonize the throat and lower gastointestinal tract in hospitalized patients[2]. *T. cutaneum* may cause white piedra in immunologically normal patients, a disease characterized by surface hard nodules along the hair shaft[3], however, skin infection by this organism is rare. *T. cutaneum* is referred to by Emmons[4] as the agent of subcutaneous and systemic fungal infection. We report a case of subcutaneous infection with *T. cutaneum* in a kidney transplanted patient.

CASE REPORT

A 32-year-old Korean woman presented with erythematous edematous patches on the right lower extremity (Fig. 1) for 2 months prior to examination in our clinic. On past history, the patient had received kidney transplantation 4 years ago due to chronic renal failure. She has received continuously cyclosporine.

A potassium hydroxide microscopic examination of the erythematous patches revealed no spores and hyphae. A biopsy specimen taken from the erythematous patches revealed dermal edema and lobular panniculitis with lymphocytic infiltration (Fig. 2). In some areas, fungal elements were noted within the subcutaneous tissue. Periodic acid-Schiff stain and Gomori methenamine silver (GMS) stain demonstrated numerous septated hyphal forms with blastoconidia in the subcutaneous tissue (Fig. 3, 5). Colonies grew rapidly in biopsied tissue cultured on Sabouraud's agar at 25℃, but incubation at 37℃ slowed the growth of colonies. The colonies were dull white or cream colored, heaped, wrinkled, waxy, and translucent (Fig. 4). Carbohydrate fermentation and assimilation of patterns revealed the organism to be *T. cutaneum*. Multiple serial blood culture failed to yield any fungal organism.

Laboratory investigations were undertaken. The hemoglobin was 9.0gm/dl; leukocyte count 22,000/mm^3 with 10% segmented neutrophils, 86% lymphocytes and 4% monocytes; peripheral blood smear showed normocytic normochromic anemia. Blood urea nitrogen/serum creatinine were 94/5.6mg/dl; the urinalysis showed protein 500mg/dl , WBC 5-9/HPF; 24 hour urine study showed protein 1.45gm/dl. Other laboratory data showed normal levels of liver function test, electrolyte, and blood sugar. On the chest X-ray, pleural effusion was seen on the right lower lung but pleural fluid culture was negative for bacteria and any fungi. An echocardiogram revealed atrial enlargement.

Based on abnormal renal function, she was treated with fluconazole. Fluconazole, daily 50mg, was given intravenously for 2 weeks

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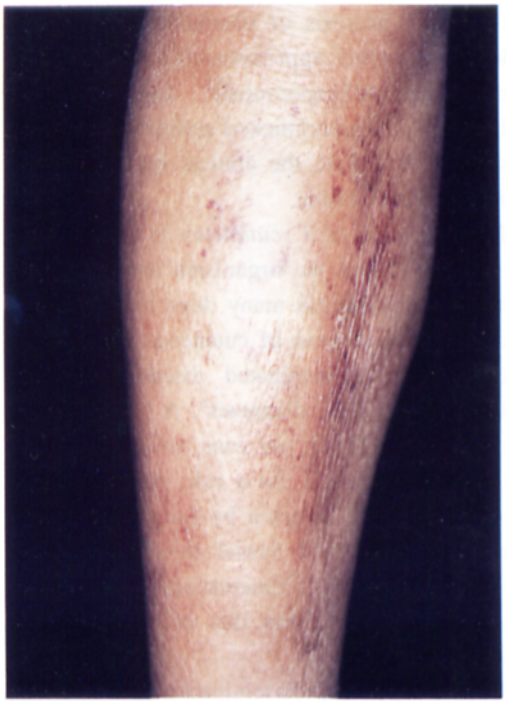

Fig. 1. Erythematous edematous patches on the right lower extremity.

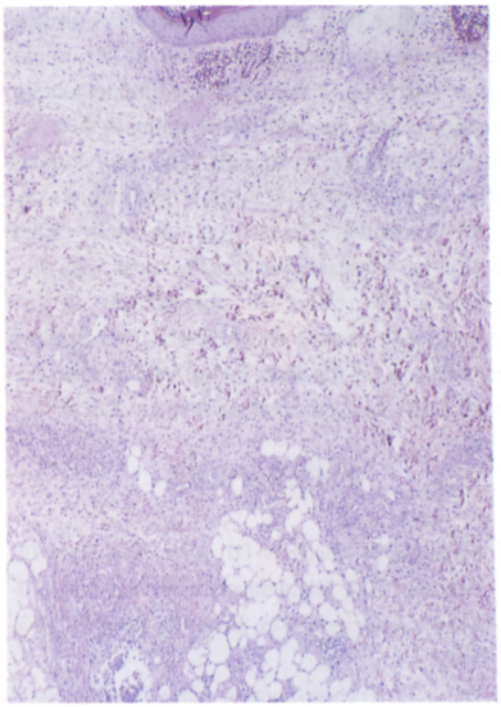

Fig. 2. Dermal edema and lobular panniculitis with lymphocytic infiltration (H & E stain, ×40).

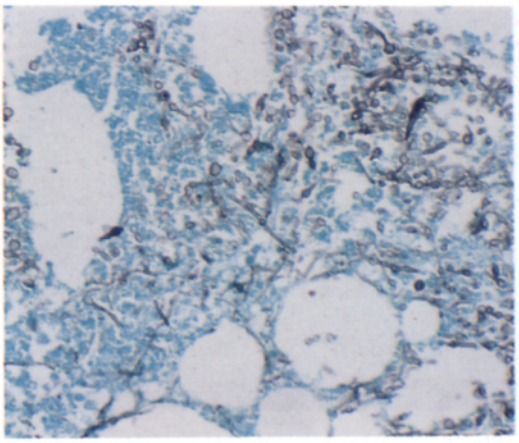

Fig. 3. Numerous hyphal forms, spores, and budding yeast cells in the subcutaneous tissue (GMS stain, ×200).

Fig. 4. The colony showed dull white or cream colored, heaped, wrinkled, waxy, and translucent in biopsied tissue cultured on Sabouraud's agar at 25℃ for 7 days.

and then given same doses orally for 8 weeks. Fungal culture in biopsy specimen after 10 weeks' treatment revealed no growth. After fluconazole were given, the cutaneous lesions gradually resolved. She remained in good skin condition at the latest follow-up, six months after the discontinuation of fluconazole.

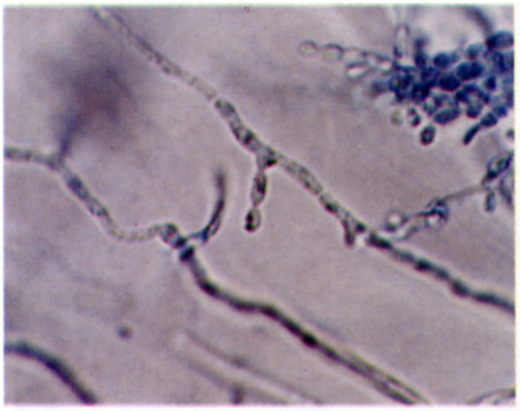

Fig. 5. True hyphae with blastoconidia produce budding yeast cells (Lactophenol cotton blue stain, ×400).

DISCUSSION

T. cutaneum has been recognized as an opportunistic pathogen that can cause potentially fatal systemic infection in immunocompromised hosts. All the reported cases of disseminated infection with *T. cutaneum* were either underlying neoplastic disease, particularly hematologic malignancies or nonneoplastic settings, including organ transplantation, intravenous drug use, acquired immune defficiency syndrome and chronic active hepatitis[2,5~10]. The present use of antimicrobial agents, irradiation, corticosteroids, immunosuppressive therapy have been associated with such an increase in opportunistic infections[11]. Our case also had suffered from chronic rejection after renal transplantation and was treated with immunosuppressive agent. In fact *T. cutaneum* is one of weakly pathogenic fungi, and cause opportunistic infections in immunocompromised hosts. The present case exemplify several risk factors, notably renal transplantation, prior antibiotics therapy and immunosuppressive therapy. With increasing experience treating immunosuppressed patients, it would be expected that more saprophytic organisms may become invasive.

The cutaneous lesions are most frequently described as purpuric papules and nodules with central necrosis or ulceration and its incidence estimated approximately 30% of patients with trichosporonosis[2]. Localized involvement of legs mimicked cellulitis has been described[12]. At first, the clinical impression of our case was cellulitis.

The diagnosis of *T. cutaneum* is usually delayed. Most often the organisms are not identified in cultures until many days later. Biopsy and culture specimens of cutaneous lesions associated with disseminated disease may be helpful for an early diagnosis[6]. A biopsy from the skin lesion, as in our case, would be helpful for the diagnosis. As a rule, the histologic finding is dermal invasion by fungal organism. Vasculitis and blood vessel thrombosis have also been reported[6]. Interestingly, histologic finding in our case was that of subcutaneous infection.

The treatment of disseminated *T. cutaneum* has been disappointing. Both amphotericin B and 5-fluorocytosine have been advocated for treatment with possible synergistic effect[13,14]. Because of the poor prognosis, alternative drugs, and other experimental methods of treatment such as interferon, leukocytes, or immunoglobulin transfusion may be justified in patients with refractory infections. In treating localized skin infection like our case, a surgical procedure may be recommended. Based on the poor renal function of our case, we tried to treat with fluconazole. The effect of fluconazole to trichosporonosis is still controversial, but marked improvement of the skin lesion of our case occurred same as a report described by Mirza[15].

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=국문초록=

신장이식술을 받은 환자에서 발생한 *Trichosporon cutaneum*에 의한 피하 감염 1예

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*Trichosporon cutaneum*은 주로 면역손상 환자에서 trichosporonosis로 알려진 국한성 또는 파종성 감염을 일으키며 피하조직과 전신성 진균감염의 원인인자로 알려져 있다. 이는 토양과 오염된 물 등에 광범위하게 존재하며 사람의 피부 및 구강 등에 정상적인 세균총으로 서식하는 *Cryptococcaceae* 계통의 진균이다.

환자는 32세 여자로 만성 신부전으로 4년전 신장 이식수술을 시행 받은 후 지속적으로 cyclosporine을 투여 받아왔으나 내원 2개월 전부터 야뇨, 빈뇨, 전신부종, 발열감, 오한 등의 증상과 함께 우측하지의 홍반성 내지 자반성의 반과 부종이 발생하였다.

병변에서 시행한 KOH 진균도말검사상 음성소견 보였으며 피부생검상 지방층염과 함께 염증세포의 침윤과 균사가 관찰되었다. 병변부 조직에서 시행한 진균 배양검사에서 25℃에서 회백색의 주름진 집락이 관찰되었으며 집락을 현미경하에서 관찰한 결과 균사가 사각형의 arthroconidia로 끊어진 모습을 관찰할 수 있었다.

이상의 소견으로 *Trichosporon cutaneum*에 의한 피하조직감염으로 진단하고 fluconazole 1일 50mg을 2주간 정맥내 투여 및 이후 8주간 경구 투여하여 피부증상의 호전을 보였다.

[의 진균지 3(1): 58-62]

색인단어: 신장이식술 (Kidney transplantation, *Trichosporon cutaneum*)