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CASE REPORT

Multifocal Systemic Sporotrichosis with Lobar Pulmonary Involvement

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Multifocal systemic sporotrichosis (disseminated sporotrichosis) with lobar pulmonary involvement is uncommon. We describe successful treatment with amphotericin B in such a patient and review data from 1 other similar case previously reported and 7 with nonlobar pulmonary involvement.

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INTRODUCTION

In the English language medical literature, 8 cases of multifocal systemic sporotrichosis with pulmonary involvement have been reported (1–8). In 7, nodular or cavitary apical lesions were present (1–6, 8). Lobar disease was present in only one (7). Our patient also had lobar disease which responded to amphotericin B treatment.

CASE REPORT

A 79-year-old man admitted to another community hospital had painful swelling on the lateral aspects of his left leg for 2 weeks, followed within 24 h by the appearance of similar lesions of his right leg, forehead, and both arms. No fever, joint pain, or cough was noted. Both tibias were tender. He had lost 3.6 kg of body weight in the preceding 4 months. The patient had worked in a gold mine at age 18 for about a year and was a casual agriculture worker. He had drunk at least one pint of hard liquor every 3 days since age 18 years.

A chest X-ray film showed infiltrate in the left lower lobe and bilateral pleural thickening (Fig. 1*a*). No bacteria or fungi were cultured from 3 sets of paired blood specimens. Serum electrophoresis showed albumin of 32 g/l and total protein of 63 g/l. The posterior tibial arteries were calcified. There was abnormal accumulation of Tc 99m in the left vertex and right parietal area of the skull, the right distal femur, the right and left proximal tibias, and the left temporomandibular joint. Microscopic examination of hematoxylin- and eosin-stained biopsies of skin lesion revealed suppurating granulomas with Langhans' giant cells but no caseation or microorganisms. However, in the methenamine-silver-stained and the periodic-acid-Schiff-stained tissues, there were oval and cigar-shaped forms.

The patient was transferred to the University of California, Davis Medical Center. The blood pressure was 118/76 mmHg, pulse was 110/min, temperature was 37.6°C, and respiratory rate was 16/min. Multiple subcutaneous nodules measuring 1–3 cm in diameter were present; they were slightly erythematous, moderately tender, and mobile. The largest, on the anterior scalp, was crusted and pustulated with multiple draining sites. The lesions on the arms and legs varied from palpable to crusting. Air movement was decreased and vocal fremitus increased over the left lower lung field posteriorly. The liver and spleen were not enlarged. The hemoglobin was 120 g/l, hematocrit was 35.5%, and the leukocyte count was 8.6×10^9/l with 9% band neutrophils. A chest X-ray film was unchanged.

Microscopic examination of previously cited stains of skin tissue from the right leg showed granulomas and vasculitis but no microorganisms. However, Sporothrix schenckii was cultured from this tissue, from sputum, and from an aspirate of the left forearm lesion. No other fungi or mycobacteria were cultured from sputum or cerebrospinal fluid. Serum in dilutions of 1:128 and 1:512, respectively, had tube and latex agglutinating antibodies to S. schenckii.

One year after a total dose of 30 mg/kg body weight of amphotericin B, the skin lesions disappeared, and a chest X-ray film (Fig. 1*b*) and 2 Tc 99m bone scans were normal.

DISCUSSION

S. schenckii is generally limited to cutaneous or subcutaneous tissue and adjacent lymphatics usually of the upper extremity. Previous reports are summarized in the accompanying table (Table I). The clinical manifestations of sporotrichosis are protean and include cutaneous lymphatic, pulmonary, extracutaneous, and disseminated forms. Tuberculosis is first suspected in nearly all patients with pulmonary involvement (9). Sarcoidosis and other fungal infections may mimic sporotrichosis or may be present concomitantly in an endemic environment. In Africa, aggressive forms of sarcoidosis affecting bone, skin, and

Table I. *Multifocal systemic sporotrichosis with pulmonary involvement*[a]*: Data summarizing previous and current cases*

N/A = unknown (not documented or available); KI = potassium iodide

Case no. (Ref.)	Age (yr)	Sex	Source of fungal isolation	Chest X-ray films[b]
1 (1)	36	F	Sputum	1 cm left apical nodule
2 (3)	61	F	Sputum, gastric washings	Fibrocalcific granulomatous disease in upper lobes, multiple cavitation
3 (4)	64	M	Lungs, pleura, at autopsy	Multiple small, noncalcified nodular densities, both apices
4 (5)	48	M	Sputum	Nodular density in left upper lobe progressing to bilateral apical cavities
5 (6)	46	M	Sputum, bronchial washings	Progression of bilateral apical infiltrates and volume loss
6 (7)	47	M	Sputum, bronchial washings	Left lower lobe consolidation
7 (8)	67	F	Nose synovium, blood cultures, sputum, gastric washings	Bilateral reticulonodular infiltrates
Current case	79	M	Sputum	Left lower lobe infiltrate and bilateral pleural thickening

[a] Associated disease: Cases 4, 5, 6 and our case had alcoholism as associated disease.
[b] Other organs involved: All cases had skin involvement, except case 4 had rectum only involved; case 5 had right knee joint only involved; case 3 had mucous membrane and joints involved in addition to the skin.
[c] Serology: Case 1 agglutination 320; case 2 positive (titer N/A); our case 128 (tube), 512 (latex).

lungs manifest clinical, radiologic, and histopathologic similarities to disseminated sporotrichosis (10).

Infections of extracutaneous tissues are rare. Of more than 3000 cases of sporotrichosis, only 5 (0.02%) reported from South Africa, deviated from the classical cutaneous and lymphangitic form; 4 of these involved extracutaneous tissue (11). In a review of 37 cases of multifocal sporotrichosis (4), the onset of illness was generally insidious; cutaneous and subcutaneous lesions, usually nodular, typically formed over the trunk, face, and extremities, and the lesions commonly became ulcerated or formed draining fistulas. Only one third had low-grade fever; rarely, chills and fever to 40°C accompanied sudden eruption of generalized skin lesions. Bone and joint pain or stiffness occurred in 13 patients. The tibia and bones of the hands and feet were most commonly affected. 11 (30%) died. Pulmonary disease may be nodular, cavitary (apical), or lobar (7) and may mimic tuberculosis, sarcoidosis, coccidioidomycosis, histoplasmosis, and blastomycosis, or a primary or metastatic neoplasm. Diagnosis is established by culture of S. schenckii from sputum, bronchial washings, pleural fluid, or tissue biopsy. Histopathologic diagnosis is difficult:

Cell-mediated immunity[c]	Therapy	Outcome
N/A	KI; total dosage N/A	Final outcome unknown
Mumps positive; histoplasmin, coccidioidin, tuberculin negative	KI, 15 drops t.i.d. initially; later amphotericin total dosage N/A	Final outcome unknown
Histoplasmin, coccidioidin, tuberculin negative	No therapy	Died before therapy
Mumps negative; histoplasmin, coccidioidin, blastomycosis negative	Amphotericin B; dosage N/A	Died (respiratory failure)
Previously positive tuberculin	Amphotericin B, 1776 mg	"Cured" at 10 months, but cough persisted
N/A	Amphotericin B, 1700 mg for 6 wks	"Cured" at 12 months
Tuberculin negative	Ketoconazole 2, 967 mg for 19 months	Recurrence, right knee
Tuberculin negative	Amphotericin B, 30 mg/kg for 8 wks	"Cured" at 1-year follow-up

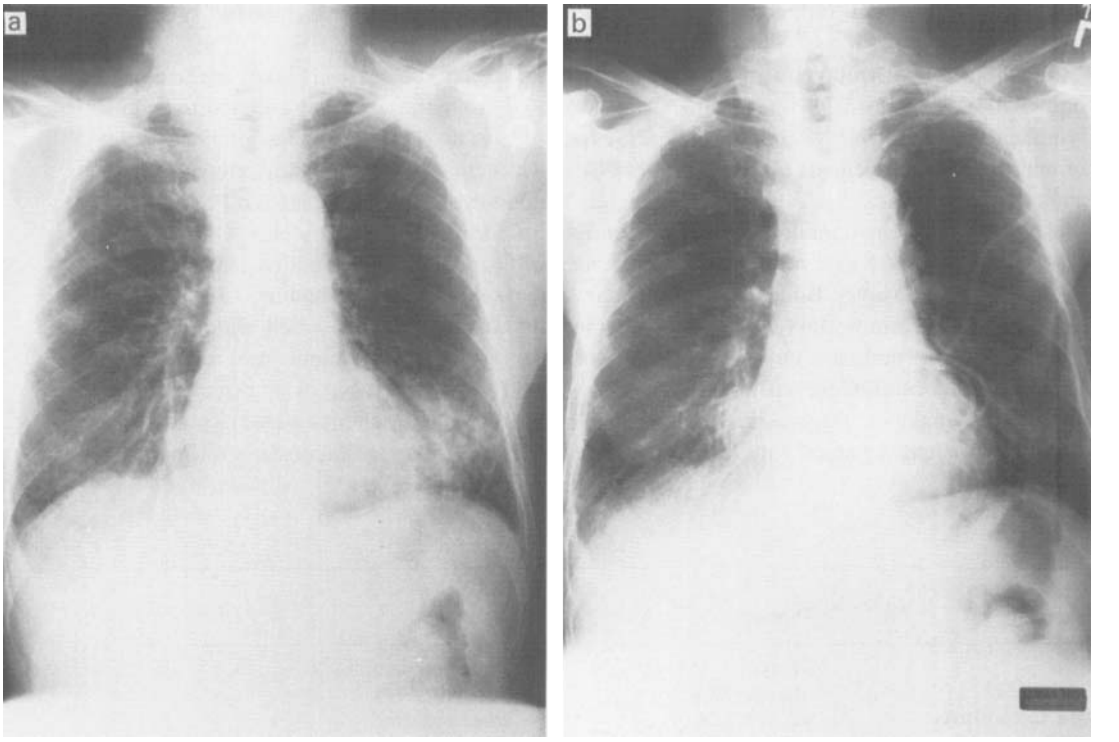

Fig. 1. Postero-anterior X-ray films of the chest: (*a*) before therapy; (*b*) during amphotericin B therapy (total dose, 20 mg/kg body weight) with Hickman catheter in place.

even with methenamine-silver and periodic acid-Schiff tissue stained fungal forms may not be found. Serology is not helpful in diagnosis or in measuring response to treatment.

In our case, bone pain was prominent in both tibias, and a bone scan showed multiple areas of abnormal uptake, although X-ray films of the tibia were normal; a follow-up bone scan 1 month after completion of therapy showed osseous abnormalities. S. schenckii was isolated from sputum, from exudate draining from skin lesions, and from punch biopsies of the skin lesions. Because there was no history of any gardening or of injuries in the preceding 2 months, the primary focus was thought to be lungs. Perhaps chronic alcoholism predisposed our patient to dissemination of S. schenckii from the lungs, even though there was an appropriate humoral antibody response.

Underlying defects of immunity have been postulated to explain the extracutaneous dissemination phenomenon (4, 11). Cell-mediated immunity was evaluated in 5 patients with cutaneous sporotrichosis and 6 patients with systemic sporotrichosis; abnormalities were noted only in the systemic group (12). We did not assess our patient's cellular immunity.

The treatment of choice in disseminated sporotrichosis continues to be amphotericin B (10). The results of susceptibility testing in vitro do not always correlate with the clinical outcome and appear to depend greatly on temperature (13).

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