
PULMONARY SPOROTRICHOSIS

IN BRAZIL: A CASE REPORT AND REVIEW

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ABSTRACT

We report a case of primary pulmonary sporotrichosis in a 74-year-old immunocompetent woman. Culture was superior to histopathological methods in terms of diagnostic yield because the organism was not abundant in tissue sections and was visualized poorly with routine histochemical stains. Ultimately, diagnosis was made by the microbiology laboratory. Recognition of this fact is important for the proper collection and transport of clinical specimens to ensure recovery of the etiological agents of mycotic infections.

KEY WORDS: *Sporothrix schenckii*; pulmonary sporotrichosis; sporotrichosis; lung.

RESUMO

Esporotricose pulmonar no Brasil. Relato de caso e revisão

Relatamos um caso de esporotricose pulmonar primária numa paciente imunocompetente com 74 anos de idade. O cultivo foi superior aos métodos histopatológicos em termos de produção diagnóstica, pois os elementos fúngicos não se encontravam em quantidade suficiente nos cortes teciduais e foram pouco visualizados nas colorações de histoquímica de rotina. Fundamentalmente, o diagnóstico foi feito pelo laboratório de microbiologia. O reconhecimento disto é importante para a adequada colheita e transporte de espécime clínico para estabelecer os agentes etiológicos de infecções micóticas.

DESCRIPTORIOS: *Sporothrix schenckii*; esporotricose pulmonar; esporotricose; pulmão.

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INTRODUCTION

Sporotrichosis is caused by the dimorphic fungus *Sporothrix schenckii* and has a worldwide distribution, especially in tropical and warm temperate climates. Most commonly the infection is acquired through traumatic implantation of the fungus present in organic material and the disease is limited to the cutaneous, subcutaneous, and lymphatic structures (15, 16). Pulmonary infection may result from inhalation of conidia into the lungs. Pulmonary sporotrichosis, a well-described disease, is being reported with increasing frequency, although in the past it has been considered a rare infection (11). In patients with acquired immunodeficiency syndrome (AIDS) the clinical features of pulmonary sporotrichosis may be different (14) to those seen in immunocompetent hosts. The disease has been observed in children (1) and there is a possibility of mixed infection. As in the case reported by Losman & Cavanaugh (7), bronchoalveolar lavage has demonstrated pulmonary co-infection with *S. schenckii* and *Pneumocystis jirovecii* (7).

This article describes the fourth Brazilian case of pulmonary sporotrichosis diagnosed by microbiological isolation of *S. schenckii* from pulmonary tissue. In addition, we review the three previously published cases.

CASE REPORT

A 74-year-old woman with a two year history of dyspnea, productive cough, low-grade fever, and malaise was referred for further evaluation. Medical history included chronic obstructive pulmonary disease (COPD) and empirical antibiotic therapy. She had no history of diabetes. At the time of admission, the patient appeared to be well nourished and had a normal physical examination with no skin lesions. This patient tested negative for human immunodeficiency virus (HIV). No predominant organism was detected in the Gram-stained sputum smears; stains for mycobacteria and fungi were negative. High-resolution computed tomography (CT) scans of the chest (Figures 1 and 2) showed diffusely distributed ground-glass attenuation, associated with traction bronchiectasis, and architectural distortion involving both lungs, without lobe predilection. The ground-glass attenuation suggested an active inflammatory process.

An open biopsy of the left lung was performed, with a wedge resection of the lingula. Lung tissue was submitted for histological studies and cultures. Examination of the biopsy specimen revealed a non-specific chronic inflammatory infiltrate, in the hematoxylin and eosin (H&E) stained tissue. The classic stain for fungi, the Gomori methenamine silver (GMS), and the standard Ziehl-Neelsen acid-fast stains were reported as negative.

Cultures of the lung tissue on Sabouraud's agar at room temperature yielded a pale mold. Microscopically, the mold-form hyphae presented delicate conidiophores bearing pyriform conidia in a rosette arrangement characteristic of *S.*

schenckii. The mold form was converted to the yeast phase at 37°C on brain-heart infusion agar. Cultures of the lung tissue for pyogenic bacteria and mycobacteria organisms revealed no growth.

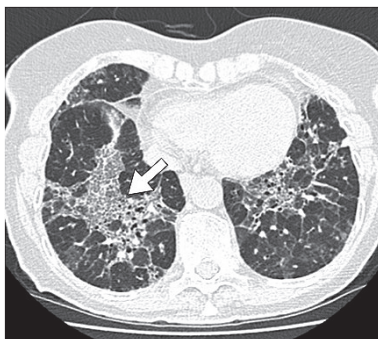


Figure 1. High-resolution CT scan of the chest shows diffusely distributed ground-glass attenuation, associated with traction bronchiectasis, and architectural distortion involving the upper lobes.



Figure 2. High-resolution CT scan of the chest shows lower lobes demonstrating similar findings and extensive ground-glass opacity (arrow). The ground-glass attenuation suggests an active inflammatory process.

The original slides from the lung biopsy were reviewed. Neither the histological reaction (H&E) nor the GMS-stained slide confirmed the etiologic diagnosis of sporotrichosis. Fungal stains were repeated and examination of multiple serial histological sections stained with GMS showed rare elongated, cigar-shaped fungal elements of *S. schenckii*.

The patient received itraconazole 400 mg daily for 6 months. Her symptoms improved, with disappearance of fever and cough. A follow-up chest CT scan demonstrated partial resolution of the lesions. The patient received itraconazole 200

mg daily for 12 additional months. No side effects were observed during itraconazole therapy and 28 months after cessation of treatment she has remained well.

Occupational history. Since it is known that plant material is the most common habitat of the fungus and that primary pulmonary sporotrichosis results from inhalation of conidia into the lung, the patient's contact with wood was investigated. She stated that she kept a greenhouse in her back yard. The disease was presumed to have been caused by inhalation of *S. schenckii* conidia during her gardening activities.

DISCUSSION

Typically, pulmonary sporotrichosis is insidious and affects men between 30 and 60 years old (13). Cough (81%) develops and is productive of sputum (66%) that may contain blood (34%). Malaise, ease of fatigue, low-grade fever (44%), and loss of weight (56%) may occur (12).

Pulmonary sporotrichosis is most often acquired as a primary infection from the inhalation of conidia. In this localized disease, for approximately 85% of reported cases, the pulmonary findings may be indistinguishable from those of secondary tuberculosis (3, 11, 17). In around 90 percent of the cases, roentgenograms of the chest revealed apical cavities, which were bilateral in nearly one third of cases (5, 12). The walls of the cavities were quite clean and the cavities contained very small amounts of caseous necrotic material. In addition, fungi other than *Aspergillus fumigatus* (4) have been reported to cause fungus balls, including *S. schenckii* (9).

Pulmonary involvement has also been described in multi-system infections (10, 11). When multiple organs are infected, most often associated with skin and joint involvement, the pulmonary pattern is quite different to that when the lungs alone are involved (3). Pulmonary sporotrichosis is usually thought to be pulmonary tuberculosis. Atypical mycobacterial infections and other pulmonary mycosis and sarcoidosis may also be confused with pulmonary sporotrichosis (10, 18).

Three cases of pulmonary sporotrichosis have been reported in Brazil (2, 6, 14). Pulmonary involvement has occurred as a primary disease in one patient with massive hemoptysis diagnosed by sputum culture (6). Multi-focal sporotrichosis with pulmonary involvement has been seen in two patients (2, 14). Other organ systems were involved in these patients, including skin and joint (2) or skin and bone (14). One of these two patients had their diagnosis confirmed by autopsy (2) and another had AIDS complicated by opportunistic sporotrichosis with the fungus present as large round elements and multiple budding cells (14).

For a definitive diagnosis of this mycosis, fungal culture should be performed. Histopathological examination is not considered the best diagnostic method for the diagnosis of sporotrichosis, because it may be difficult to identify *S. schenckii* in histological sections stained with routine histochemical stains such as H&E and GMS (8).

In conclusion, the diagnosis of pulmonary sporotrichosis requires a high level of clinical suspicion and awareness of the tests commonly used to identify fungal diseases, including special stains in histopathological studies of tissue and cultures. These tests vary in sensitivity in different clinical findings. Each has certain limitations that must be recognized if they are to be used correctly, in order to be able to interpret their results.

REFERENCES

1. Callens SF, Kitelete F, Lukun P, Lelo P, Van Rie A, Behets F, Colebunders R. Pulmonary *Sporothrix schenckii* infection in a HIV positive child. *J Trop Pediatr* 52: 144-146, 2006.
2. Carneiro JF. Micoses pulmonares no Brasil. *Rev Serv Nac Tuberc* 4: 183-210, 1960.
3. Comstock C, Wolson AH. Roentgenology of sporotrichosis. *Am J Roentgenol Radium Ther Nucl Med* 125: 651-655, 1975.
4. Guazzelli LS, Severo CB, Hoff LS, Pinto GL, Camargo JJ, Severo LC. *Aspergillus fumigatus* fungus ball in the pleural cavity. *J Bras Pneumol* 38: 125-132, 2012.
5. Kauffman CA, Bustamante B, Chapman SW, Pappas PG. Clinical practice guidelines for the management of Sporotrichosis: 2007 update by the Infectious Diseases Society of America. *Clin Infect Dis* 45: 1255-1265, 2007.
6. Lacaz CS. Diagnóstico e tratamento das micoses pulmonares. *Rev Paul Med* 38: 389-400, 1951.
7. Losman JA, Cavanaugh K. Cases from the Osler Medical Service at Johns Hopkins University. *Am J Med* 117: 353-356, 2004.
8. Marques MEA, Coelho KIR, Sotto MN, Bacchi CE. Comparison between histochemical and immunohistochemical methods for diagnosis of sporotrichosis. *J Clin Pathol* 45: 1089-1093, 1992.
9. Mohr JA, Patterson CD, Eaton BG, Rhoades ER, Nichols NB. Primary pulmonary sporotrichosis. *Am Rev Respir Dis* 106: 260-264, 1972.
10. Palomino J, Saeed O, Daroca P, Lasky J. A 47-year-old man with cough dyspnea, and an abnormal chest radiograph. *Chest* 135: 872-875, 2009.
11. Pluss JL, Opal SM. Pulmonary sporotrichosis: review of treatment and outcome. *Medicine (Baltimore)* 65: 143-1453, 1986.
12. Powel KE. Sporotrichosis. In: Eisenstein HE, editor. *Handbook on fungal diseases*. Illinois, American College of Chest Physicians; 1981. p. 75-83.
13. Ramos-e-Silva M, Vasconcelos C, Carneiro S, Cestari T. Sporotrichosis. *Clin Dermatol* 25: 181-187, 2007.
14. Rocha MM, Dassin T, Lira R, Lima EL, Severo LC, Londero AT. Sporotrichosis in patient with AIDS: report of a case and review. *Rev Iberoam Micol* 18: 133-136, 2001
15. Rosa ACM, Scroferneker ML, Vettorato R, Gervini RL, Vettorato G, Weber A. Epidemiology of sporotrichosis: A study of 304 cases in Brazil. *J Am Acad Dermatol* 52: 451-459, 2005.
16. Schubach A, Barros MB, Wanke B. Epidemic sporotrichosis. *Curr Opin Infect Dis* 21: 129-133, 2008.
17. Singhai M, Rawat V, Verma P, Jha PK, Shree D, Goyal R, Umesh. Primary pulmonary sporotrichosis in a sub-Himalayan patient. *J Lab Physicians* 4: 48-49, 2012.
18. Yang DJ, Krishnan RS, Guillen DR, Scchmiege LM, Leis PF, Hsy S. Disseminated sporotrichosis mimicking sarcoidosis. *Int J Dermatol* 45: 450-453, 2004.