

Sporotrichosis in patient with AIDS: report of a case and review

Marineide M. Rocha¹, Terezinha Dassin¹, Rita Lira¹, Eduardo L. Lima², Luiz Carlos Severo³ & Alberto T. Londero⁴

Serviços de ¹Infectologia e de ²Anatomia Patológica, Hospital Nossa Senhora da Conceição, Porto Alegre; ³Universidade Federal do Rio Grande do Sul, Porto Alegre; ⁴Universidade Federal de Santa Maria, Santa Maria, RS, Brasil

Summary

Although sporotrichosis is not an AIDS-defining infection, reports of sporotrichosis in individuals infected with HIV are increasing. We report an unusual case of this co-infection in a man with progressive deep cutaneous ulcerations with numerous pleomorphic yeast cells of *Sporothrix schenckii*. In addition a review of the literature on this subject was carried out and commented upon.

Key words

Sporotrichosis, *Sporothrix schenckii*, HIV, AIDS

Esporotricosis en un paciente con sida: descripción de un caso y revisión

Resumen

Si bien la esporotricosis no es considerada una enfermedad que defina al sida, se ha comprobado un aumento del número de casos en pacientes VIH positivos. Presentamos un caso raro de esta asociación mórbida, en un hombre con úlceras cutáneas profundas y progresivas, en las que se encontraron microscópicamente numerosas células levaduriformes, esféricas o pleomorfas de *Sporothrix schenckii*. La literatura sobre este tema fue revisada y comentada en el trabajo.

Palabras clave

Esporotricosis, *Sporothrix schenckii*, VIH, Sida

Sporothrix schenckii is a thermal dimorphic pathogenic fungus that, due to where and how it lives in the environment, may usually be implanted through the skin causing a subcutaneous mycosis. More rarely it produces propagules that may be dispersed by the air and inhaled by a host causing clinical manifestations similar to those of the other thermal dimorphic pathogenic fungi, agents of systemic mycosis [1]. The similarities between the systemic manifestation of sporotrichosis and those of the systemic mycosis was emphasized by Lynch, in 1970, who also recognized the role of *S. schenckii* as an opportunistic invader [2].

The opportunistic form of sporotrichosis occurs by implantation or by inhalation of the propagules of *S. schenckii* by an immunocompromised host. Many predisposing conditions of the host have been pointed out: alcoholism, diabetes mellitus, hematologic malignancies, chronic obstructive pulmonary disease, long term treat-

ment with corticosteroids, transplant recipients and patients with AIDS [3].

In this paper we will describe a case of opportunistic sporotrichosis in a patient with AIDS. The literature will be reviewed and some aspects of the published cases are also presented.

CASE REPORT

A 29 year-old man with a history of alcoholism, intravenous drug use and HIV infection was admitted to the emergency room of the hospital in a very bad condition. He presented skin lesions since six months ago and fever, dysphagia and hoarseness since three months ago.

Myriad of small and many large skin lesions over the patient's body were observed. Large scale crusted infiltrative lesions with irregular borders were distributed on his face, forehead and nose (Figure 1); many acneiform or furuncle-like lesions, some ulcerated, were scattered over the thorax. Ulcerated lesions in the right upper limb, one of them associated with an ascendant chain of nodules, were observed. Two large phagedenic ulcers with delimited borders and some smaller ulcerations were distributed on the lower limbs. A very large ulcerated lesion with purulent basis extended from the perineal region to the scrotum (Figure 2). Oral candidosis and small ulcers were seen in the mouth.

Laboratory findings: hemoglobin 9 g/dl, hematocrit 27.2%; blood leukocytes count 8.6×10^3 (24% lymphocytes, 13% band forms and 55% polymorphonuclear cells); ALT 44 U/L, ASF 131 U/L; alkaline phosphatase 373 U/L; other biochemical examinations in normal values. His CD4 count was $228/\text{mm}^3$ with 10^4 HIV RNA copies/ml. Chest X-ray revealed an interstitial infiltration

Dirección para correspondencia:

Dr. Luiz Carlos Severo
Laboratório de Microbiologia Clínica,
IPD - Santa Casa, Annes Dias,
285 Porto Alegre 90020-090
RS Brazil
Fax: +55 513 214 8435
E-mail: severo@santacasa.tche.br

Aceptado para publicación el 15 de Junio de 2001



Figure 1. Infiltrative lesions with irregular borders on face, forehead and nose.

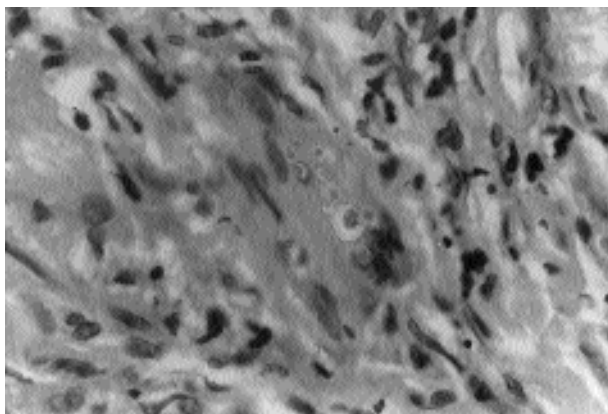


Figure 3. Dermal granuloma from lesion biopsy seen in figure one. Many yeast cells are seen within a large giant cell (H&E, x 250).



Figure 2. Ulcerated lesions with purulent basis in scrotum and left lower limb.

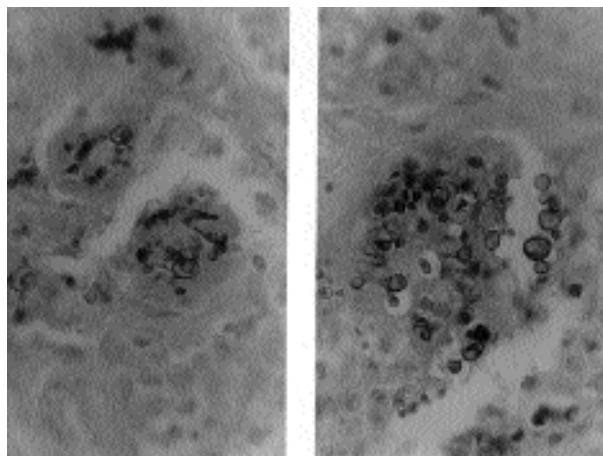


Figure 4. *Sporothrix schenckii* (Replicate section of figure 3) illustrate the variation in shape and in size of the buds (GMS, x 250).

and diffuse small alveolar consolidations in both lungs. Arterial gasometry did not reveal hypoxia. An X-ray of the left forearm showed a cystic lesion on distal ulnar extremity.

A biopsy of the skin lesion and an aspirate of an abscess were performed and an empiric treatment was initiated with amphotericin B, erythromycin, gentamicin, oxacillin and acyclovir.

Skin biopsied specimen stained by haematoxylin and eosin (H&E) showed suppurative granulomatous dermatitis with many eosinophils and multinucleated giant cells (Figure 3), but stained by Gomori-Grocott technique (GMS) revealed many small pleomorphic ovoid, cigar-shaped budding yeast, and multiple budding cells (Figure 4). Characteristic colonies of *Sporothrix schenckii* were obtained in cultures of the pus and fragments of the biopsied tissue.

After a very short period of improvement, patient's respiratory conditions worsened abruptly. Sputum examination revealed many Gram-negative bacilli and *Klebsiella oxytoca* was isolated in culture. The patient died ten days after his admission. No autopsy was permitted.

DISCUSSION

Sporotrichosis has been an uncommon opportunistic infection in AIDS patients. Twenty reported cases could be gathered in the literature [3-22] and a new one is herein related. The mycosis may be the presentation of AIDS [3-7], but more frequently it occurred in patients with already recognized HIV infection [8-22, present report]. It affected patients aged 20 to 71 years (mean 39 years) and more frequently males (18) than females (3). With the exception of one patient with invasive sinusitis [16] and another one with pulmonary infection [12], the remaining 19 patients presented skin lesions at the presentation of the mycosis. Usually cutaneous lesions were multiple and widespread. Initially and later on, clinical manifestations as well as the involved organ at the time of the diagnosis and later on are shown in table 1.

Diagnosis of 16 out of 18 patients presenting cutaneous lesions was accomplished by the examination of specimens obtained by histology [3,4,9,15] isolation in culture [14] or both, histology and culture [5,7,8,10,11,18,20,22, present report] or pus aspirated from an abscess - microscopy [13] or culture [6]; the remaining two

Table 1. Clinical manifestations and involved sites or organs in HIV patients with sporotrichosis.

| Ref. | Clinical manifestations | | Specimen/organ where the fungus was detected | |
|------|--|---|--|--|
| | Initially | Later on | At diagnosis | Later on |
| [3] | Skin lesions | Arthritis, osteomyelitis | Skin | Skin, blood |
| [4] | Skin lesions | - | Skin | Skin, sputum, anterior arm |
| [5] | Skin lesions, cough, weight loss | Ocular lesions, arthritis, seizure | Skin, blood, bone marrow, sputum | Skin*, lymph node*, lung*, esophagus*, colon patela*, testes*, synovium, knee* |
| [6] | Skin lesions | - | Skin | Skin |
| [7] | Skin lesions | - | Skin | Skin |
| [8] | Skin lesions | Arthritis | Skin | - |
| [9] | Skin lesions | Respiratory infection | Skin | Skin, sputum |
| [10] | Skin lesions | Infiltrative lesions on the cheek, seizures | Skin, brain, spinal fluid | Skin*, lung*, prostate*, leptomeninges*, meninges* |
| [11] | Skin lesions | - | Skin | - |
| [12] | Respiratory infection | - | Sputum | Skin |
| [13] | Skin lesions | - | Skin | Skin |
| [14] | Skin lesions | Endophthalmitis | Skin | Aqueous and vitreous tap |
| [15] | Skin lesions, arthritis | Encephalitis | Skin, joint fluid | Skin, joint fluid, lung*, liver*, spleen* |
| [16] | Sinusitis | - | Surgical specimen | - |
| [17] | Skin lesions, arthritis | - | Joint fluid | - |
| [18] | Skin lesions | Skin | Cerebrospinal fluid | Cerebrospinal fluid |
| [19] | Skin lesions, headache | - | Skin, urine, cerebrospinal fluid | Meninges*, brain*, lung* |
| [20] | Skin lesions, arthritis | - | Skin | Synovial |
| [21] | Skin lesions, arthritis | Testis | Joint fluid, blood | Skin |
| [22] | Skin lesions, arthritis, lymphadenopathy | Tuberculosis | Skin, joint fluid, blood | - |
| PR | Skin lesion | - | Skin | -- |

* Autopsy finding; PR, present report

patients [17,21] culture of aspirated joint fluid led to diagnosis of the mycosis. Surgical specimen and sputum were used for the diagnosis of the patients presenting, respectively, sinusitis [16] and pulmonary infection [12] at the presentation of the mycosis.

In cut sections or smeared pus *S. schenckii* was recognised by the peculiar cigar-shaped yeast form; however, in two patients [4,9] the fungus was present as large round elements reaching more than 8 µm in diameter, simulating *Cryptococcus neoformans*, agent of a more frequent opportunistic infection in AIDS patients.

Al-Tawfiq *et al.* [3] revised the treatment of sporotrichosis in AIDS patients pointing amphoterecin B as the drug of choice for initial use and itraconazole as maintenance therapy. This recommendation is in accordance with practice guidelines for the management of patients with sporotrichosis and AIDS for the Mycoses Study Group, Infectious Diseases Society of America [23].

Finally, our patient has an intermediate-stage HIV infection, based on the CD4 count (228/mm³) and viral copies (10⁴/ml). We may presume that the disseminated sporotrichosis was facilitated by his alcohol abuse [24].

References

- Wilson JW. Sporotrichosis – The importance and significance of the portal of inoculation and the chancreiform syndrome. In: Clinical and immunologic aspects of fungus disease. Springfield, Charles C Thomaz, 1957: 86-96.
- Lynch PJ, Voorhees JJ, Harrel R. Systemic sporotrichosis. Ann Intern Med 1970; 73: 23-30.
- Al-Tawfiq JA, Wools KK. Disseminated sporotrichosis and *Sporothrix schenckii* fungemia as the initial presentation of human immunodeficiency virus infection. Clin Infect Dis 1998; 26: 1403-1406.
- Fitzpatrick JE, Eubanks S. Acquired immunodeficiency syndrome presenting as disseminated cutaneous sporotrichosis. Int J Dermatol 1988; 27: 406-407.
- Heller HM, Fuhrer J. Disseminated sporotrichosis in patients with AIDS: case report and review of the literature. AIDS 1991; 5: 1243-1246.
- Matter SE, Bailey DM, Sexton DJ. Immune deficiency presenting as disseminated sporotrichosis. J Oklahoma State Med Assoc 1984; 77: 114-117.
- Neto RJP, Machado AA, Castro G, Quaglio ASS, Martinez R. Esporotricose cutânea disseminada como manifestação inicial da síndrome da imunodeficiência adquirida: relato de caso. Rev Soc Bras Med Trop 1999; 32: 57-61.
- Bibler MR, Luber HJ, Glueck HI, Estes SA. Disseminated sporotrichosis in a patient with HIV infection after treatment for acquired factor VIII inhibitor. JAMA 1986; 256: 3125-3126.
- Bolao F, Podzamczar D, Ventin M, Gudiol F. Efficacy of acute phase and maintenance therapy with itraconazole in an AIDS patient with sporotrichosis. Eur J Clin Microbiol Infect Dis 1994; 13: 609-6120.
- Donabedian H, O'Donnell E, Olszenwski C, MacArthur RD, Budd N. Disseminated cutaneous and meningeal sporotrichosis in an AIDS patient. Diagn Microbiol Infect Dis 1994; 18: 111-115.

11. Goldani LZ, Aquino VR, Dargél AA. Disseminated cutaneous sporotrichosis in an AIDS patient receiving maintenance therapy with fluconazole for previous cryptococcal meningitis. *Clin Infect Dis* 1999; 28: 1337-1338.
12. Gori S, Lupetti A, Moscato G, Parenti M, Lofaro A. Pulmonary sporotrichosis in a human immunodeficiency virus - infected patient. A case report. *Acta Cytol* 1997; 41: 519-520.
13. Keiser P, Whittle D. Sporotrichosis in human immunodeficiency virus-infected-patients: report of a case. *Rev Infect Dis* 1991; 13: 1027-1028.
14. Kurosawa A, Pollock SC, Collins MP, Kraff CR, Tso MO. *Sporothrix schenckii* endophthalmitis in a patient with human immunodeficiency virus infection. *Arch Ophthalmol* 1988; 106: 376-380.
15. Lipstein-Kresh E, Isenberg HD, Singer C, Cooke O, Greenwald RA. Disseminated *Sporothrix schenckii* infection with arthritis in a patient with acquired immunodeficiency syndrome. *J Rheumatol* 1985; 12: 805-808.
16. Morgan M, Reves R. Invasive sinusitis due to *Sporothrix schenckii* in patient with AIDS. *Clin Infect Dis* 1996; 23: 1319-1320.
17. Oscherwitz SL, Rinaldi MG. Disseminated sporotrichosis in a patient infected with human immunodeficiency virus. *Clin Infect Dis* 1992; 15: 568-569.
18. Penn CC, Goldstein E, Bartholomew WR. *Sporothrix schenckii* in a patient with AIDS. *Clin Infect Dis* 1992; 15: 741-743.
19. Rotz LD, Slater LN, Wack MF, Boyd AL, Scott EN, Greenfield RA. Disseminated sporotrichosis with meningitis in a patient with AIDS. *Infect Dis Clin Practice* 1996; 5: 566-568.
20. Shaw JC, Levinson W, Montanaro A. Sporotrichosis in the acquired immunodeficiency syndrome. *J Am Acad Dermatol* 1989; 21: 1145-1147.
21. Ware AJ, Cockerell CJ, Skies DJ, Kussman HM. Disseminated sporotrichosis with extensive cutaneous involvement in a patient with AIDS. *J Am Acad Dermatol* 1999; 40: 350-355.
22. Edwards C, Reuther WL III, Greer DL. Disseminated osteoarticular sporotrichosis: treatment in a patient with acquired immunodeficiency syndrome. *South Med J* 2000; 93: 803-806.
23. Kaufman CA, Hajjeh R, Chapman SW. (USA, Mycoses Study Group). Practice guidelines for the management on patients with sporotrichosis. *Clin Infect Dis* 2000; 30: 684-687.
24. Wang JP, Granlund KF, Bozzette SA, Botte MJ, Fierer J. Bursal sporotrichosis: case report and review. *Clin Infect Dis* 2000; 31: 615-615.