

Disseminated sporotrichosis in a human immunodeficiency virus positive man with upper airway involvement

Xian Fui Tan, MD¹, Wee Fu Gan, MRCP¹, Nor Zaila Zaidan, MIntMed¹, Syziah Ibrahim, Path²

¹Infectious Disease Unit, Internal Medicine Department, Hospital Melaka, Melaka, Malaysia, ²Pathology Department, Hospital Melaka, Melaka, Malaysia

SUMMARY

Sporotrichosis is a fungal infection caused by dimorphic fungus *Sporothrix*. Sporotrichosis can be seen in cutaneous or extracutaneous form. Disseminated form of sporotrichosis with upper airway involvement rarely been reported. We present a case of disseminated sporotrichosis in a HIV-positive young gentleman manifesting as fungemia, lymphocutaneous and upper airway involvement.

INTRODUCTION

Sporotrichosis is a fungal infection caused by dimorphic fungus *Sporothrix*, which is found in decaying vegetation, sphagnum moss, soil, and other environmental niches throughout the world.^{1,3} Transmission involves subcutaneous inoculation through injury from thorns or animal bites or scratches. It commonly afflicts humans in the lymphocutaneous form. Rarely it might be disseminated involving the lungs, osteoarticular or central nervous system. Here we present a case of a disseminated sporotrichosis in a HIV-positive young gentleman manifesting as fungemia, lymphocutaneous and upper airway involvement.

CASE REPORT

A 24-year-old gentleman presented with diffuse skin lesions and fever for one month. He was newly diagnosed HIV-positive with a CD4 count of 26 and was not on any anti-retroviral (ARV) medication yet. He is working as an administration officer, has no pet and volunteered no history of gardening or exposure to soil. On physical examination there were multiple ulcerous and crusted nodular lesions over the face, bilateral upper and lower limbs along the lymphatic channel, as well as numerous firm, non-tender lymph nodes at the submandibular, submental, and supraclavicular regions. He also had multiple interphalangeal joints swelling over bilateral hands and feet where the x-rays revealed osteopenia over involved joints (Figure 1). Despite treatment for candidiasis, he had persistent dysphagia and odynophagia whereby intraoral inspection revealed many small ulcerated superficial lesions measuring less than 0.5cm each, on the soft palate. During endoscopy, generalized lobulated lesions were also seen in the nasal cavity, laryngeal surface, epiglottis, bilateral arytenoid, and pyriform fossa (Figure 2). Otherwise, he had no respiratory symptoms and had clear chest x-ray.

Provisional diagnosis of systemic fungal infection was entertained and intravenous amphotericin B deoxycholate

initiated empirically. Subsequent blood fungal culture grew *Sporothrix schenckii*. Lactophenol cotton blue (LPCB) mount of slide culture demonstrated small budding, round, oval-or-cigar shaped, 2-6µm diameter yeast cells, which is typical of *Sporothrix species*. Histopathology examination of both skin and nasal biopsies also revealed sporotrichosis (Figure 3). The organism was later sent for antifungal susceptibility test and was reported sensitive to Amphotericin B and itraconazole.

The patient responded clinically to the intravenous Amphotericin B with dosage of 1mg/kg/dose for total 14 days with an improvement of skin lesions (Figure 5) and lymphadenopathy, interphalangeal joint swelling also subsided. Subsequent repeated blood fungal culture was negative and inflammatory markers were much improved. Upon discharge, antiretroviral therapy was commenced, and intravenous Amphotericin B was transitioned to oral itraconazole 200mg 3 times per day, treatment was intended till his CD4 count more than 200 cells /uL or HIV viral load suppressed. He remained well during his clinic appointment with further resolution of cutaneous lesions and residual lymphadenopathy.

DISCUSSION

Sporotrichosis is a disease caused by a thermo-dimorphic fungus *Sporothrix species*.^{1,3} It is currently reported throughout the world, especially in tropical and subtropical regions as the humid environment favours the fungal growth. It grows as mold in the environment and at temperature below 35°C and assumes the yeast form at temperature above 35°C.² The commonest route of transmission is by subcutaneous inoculation of the organism through traumatic inoculation⁴, and occasionally inhalation of conidia causing upper airway and pulmonary infection.

Sporotrichosis can generally be classified into cutaneous or extracutaneous form.^{1,2} The cutaneous form can manifest as either cutaneous fixed type or lymphocutaneous type, which are both found predominantly in the immunocompetent host.^{1,2} Extracutaneous form is rare and usually in immunocompromised patients.⁵ It can be divided into disseminated cutaneous type, disseminated or pulmonary sporotrichosis. Laryngeal and nasal sporotrichosis are rarely described. We illustrated here a patient with advanced HIV who was treated for disseminated sporotrichosis involving the nasal, oral, laryngeal, lymphocutaneous and hematogenous systems. In a patient with advanced HIV, the immunocompromised state of the body might aggravate the

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Corresponding Author: Xian Fui Tan

Email: xianfui93@gmail.com



Fig. 1: X-rays osteopenia in all involved joints over bilateral hands and feet.

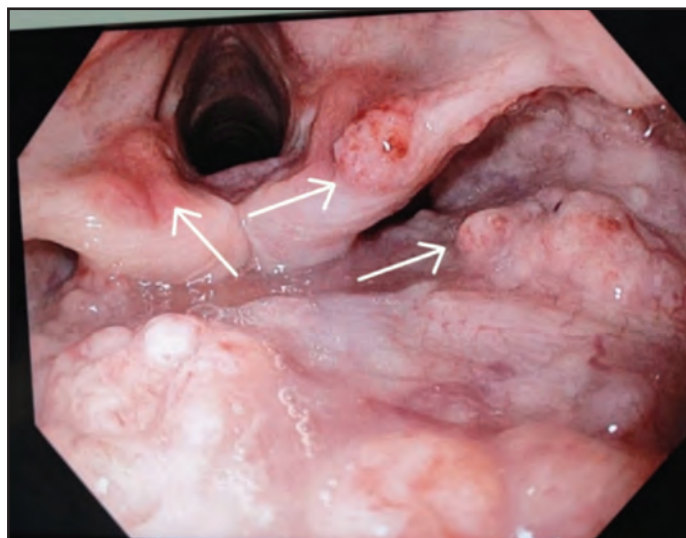


Fig. 2: Endoscopy findings of multiple lobulated erythematous lesions over laryngeal surface, epiglottis and bilateral arytenoid.

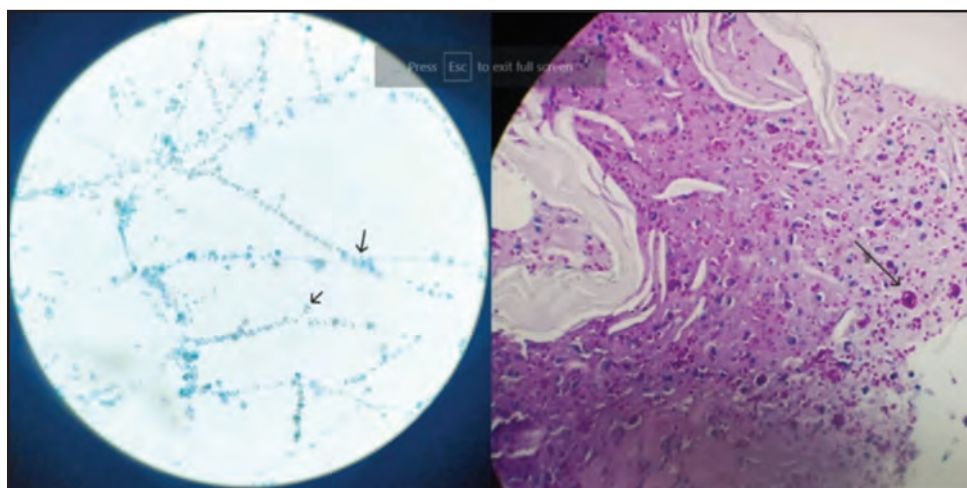


Fig. 3: (Left): Microscopy shows small budding, round, oval-or-cigar shaped, with 2-6µm diameter yeast cells in LPCB mount of slide culture.
 (Right): Densely eosinophilic yeast forms with a surrounding ray of eosinophilic material called 'Sporothrix asteroids' seen in the histopathology examination.

sporotrichosis, with higher incidences of severe disseminated form of the disease. Hence, the diagnosis of cutaneous form of sporotrichosis in an immunocompromised host should spark a search for dissemination to the other sites, especially upper airway, as although uncommon, sporotrichosis can be transmitted by inhalation.

So far, we are only aware of two case reports describing laryngeal involvement in adults. Roslle N. described an immunocompetent individual with hoarseness due to laryngeal sporotrichosis.⁶ Also described in the literature a HIV positive man who presented with painful intraoral papular-infiltrative lesions and dysphagia, and subsequently found to have laryngeal sporotrichosis.⁷

Although upper airway involvement is rare in sporotrichosis, it is important for clinicians to be aware of the possibility. Among people living with HIV, complaints of odynophagia or dysphagia are often treated as oral candidiasis. However, other diagnosis should not be overlooked and sporotrichosis should also be considered in the evaluation of an immunocompromised patient including newly diagnosed advance RVD.

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CONFLICTS OF INTEREST

The authors declare that they have no conflict of interest.

REFERENCES

1. Kontoyiannis DP, Slavin M. Fungal infections in immunocompromised hosts. 2019; :121-43.
2. Barros M, de Almeida Paes R, Schubach A. Sporothrix schenckii and Sporotrichosis. Clinical Microbiology Reviews. 2011; 24(4): 633-654.
3. Zancopé Oliveira RM, de Almeida Paes R, Hagen F. Urban environment. Environmental Mycology in Public Health. 2016; :147-55.
4. Mahajan V. Sporotrichosis: An Overview and Therapeutic Options. Dermatology Research and Practice. 2014; 2014: 1-13.
5. Saeed L, Weber RJ, Puryear SB, Bahrani E, Peluso MJ, Babik JM, et al. Disseminated cutaneous and osteoarticular sporotrichosis mimicking pyoderma gangrenosum. Open Forum Infectious Diseases. 2019; 6(10).
6. Roslle N, Noh KB, Ahmad MA, Sachlin IS, Husain S. Nasal and laryngeal sporotrichosis in an immunocompetent individual: A case report. The Egyptian Journal of Otolaryngology. 2023; 39(1).
7. Campos Fontes P, Kitakawa D, Rodarte Carvalho Y, Brandão A, Guimarães Cabral L, Almeida J. Sporotrichosis in an HIV-Positive Man with Oral Lesions. Acta Cytologica. 2007 ;51(4): 648-650.