

Refractory sporotrichosis lesion: An effective and pioneering approach in a patient living with human immunodeficiency virus/acquired immunodeficiency syndrome



Rodrigo do Carmo Silva, MD,^a Dayvison Francis Saraiva Freitas, MD, PhD,^b João Pedro Cabrera Pereira, MD,^a Antonio Carlos Francesconi do Valle, MD, PhD,^b Maria Clara Gutierrez-Galhardo, MD, PhD,^b Fernando Almeida-Silva, PhD,^b and Felipe Mauricio Soeiro Sampaio, MD, MSc^a

Key words: curettage; electrosurgery; itraconazole; *Sporothrix*; sporotrichosis.

INTRODUCTION

Sporotrichosis is a subcutaneous mycosis caused by fungi of the genus *Sporothrix*.¹ Since 1998, in the metropolitan region of Rio de Janeiro, there has been an increase in the number of human cases, in a zoonotic transmission pattern, from domestic and stray cats, involving *Sporothrix brasiliensis* as the causative species.¹

The primary treatment for this infection includes oral itraconazole, potassium iodide, terbinafine, and intravenous amphotericin B.¹ Refractory cases and the cases in which the patients have contraindications to pharmacological treatment may benefit from adjuvant methods such as cryosurgery, thermotherapy, and electrosurgery.²

Here, we report a case of lymphocutaneous sporotrichosis refractory to itraconazole treatment that achieved clinical cure after electrosurgery with curettage.

CASE REPORT

A 52-year-old male patient reported the development of skin lesions on the dorsum of his left foot after a cutting trauma with a piece of glass. The lesions showed an ascending progression to the medial surface of the left leg (Fig 1).

He was diagnosed with human immunodeficiency virus infection 1 year before and was receiving regular antiretroviral treatment with tenofovir, lamivudine, and efavirenz. The T-CD4+ lymphocyte count was 809 cells/mm³, and the viral load was below 40 copies/mL. No other comorbidities were noted.

Dermatological examination revealed 3 similar erythematous, oval, and well-delimited plaques (1–3 cm in diameter) on the dorsum of his left foot and erythematous nodules on the left leg. The plaques were centered by ulcerations covered by black crusts.

A biopsy was performed; however, the results were inconclusive, showing pseudoepitheliomatous squamous hyperplasia and a nonspecific acute and chronic inflammatory process. The culture of the fragment of a skin lesion isolated *Sporothrix* spp.; therefore, treatment was initiated with itraconazole 200 mg/day. During month 7 of treatment, the leg lesions healed; however, there were still active lesions on the foot. Therefore, daily thermotherapy (about 40–45 °C, three times a day for 20 minutes) was combined for 3 months to cure the remaining active lesions unsuccessfully. This was followed by 10 monthly sessions of cryosurgery using liquid

From the Department of Dermatology, Bonsucesso Federal Hospital, Rio de Janeiro, Brazil^a; Evandro Chagas National Institute of Infectious Diseases, Oswaldo Cruz Foundation, Rio de Janeiro, Brazil.^b

Funding sources: None.

IRB approval status: This case report is part of a cohort study approved by the Evandro Chagas National Institute of Infectious Diseases IRB under the appreciation number 88551018.9.0000.5262.

Patient consent: The patient gave consent for his photographs and medical information to be published in print and online and with the understanding that this information may be publicly available and anonymized. Consent for publication was

obtained by the authors and proof of consent is available upon request.

Correspondence to: Rodrigo do Carmo Silva, MD, Av. Londres, 616, Bonsucesso, Rio de Janeiro, Rio de Janeiro, Brazil, 21041-020. E-mail: dr.rodrigodocarmo@gmail.com.

JAAD Case Reports 2022;26:32-4.

2352-5126

© 2022 by the American Academy of Dermatology, Inc. Published by Elsevier, Inc. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

<https://doi.org/10.1016/j.jidcr.2022.05.040>



Fig 1. Lymphocutaneous sporotrichosis: Three erythematous, well-delimited plaques, centered by ulceration covered by crusts, 1 to 3 cm in diameter, on the dorsum of the left foot.



Fig 2. Lymphocutaneous sporotrichosis on the left lower limb: Preoperative.

nitrogen spray (each session included two 15- to 30-second cycles, 1 minute apart, using an open tip B - CryAc Brymill). However, there was only partial improvement in the condition of the patient. As the patient had discomfort with the pain caused by cryosurgery sessions and nausea caused by itraconazole, an electrosurgery with curettage was indicated after 25 months of pharmacological treatment.

Antisepsis was performed using digluconate and alcoholic chlorhexidine; local anesthesia was administered using 2% lidocaine, 1:1000 epinephrine, and 0.9% saline solution. The lesion curettage was performed using a curette number 02, and the electrosurgery was performed using an electrocoagulation knife tip (biterminal electric scalpel, with monopolar pen WEN HF 120) (Figs 2 and 3). A dressing was applied daily, after cleaning the wound using 0.9% saline, followed by application of 1% silver sulfadiazine cream, and covering it with gauze.

The wound was healed by secondary intention in less than 2 months after the electrosurgery, and then itraconazole was discontinued. After 2 months, the procedure left a scar that was initially hyperpigmented on the periphery with a slightly erythematous



Fig 3. Lymphocutaneous sporotrichosis on the left lower limb: Intraoperative, after curettage.

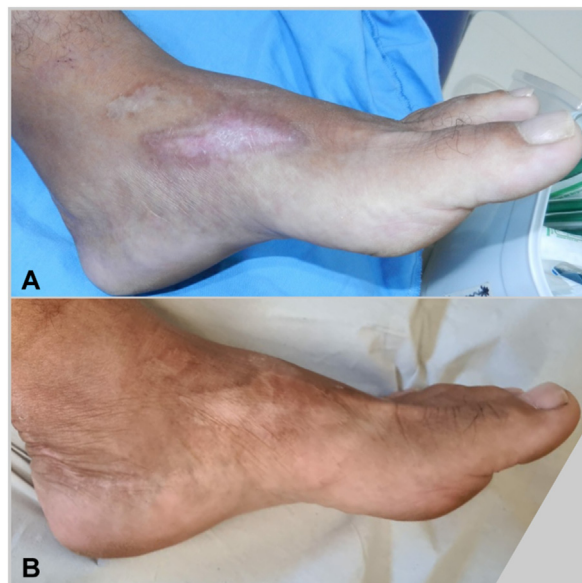


Fig 4. Lymphocutaneous sporotrichosis on the left lower limb: Scar after 2 months (A) and after 3 years (B) of the electrosurgery with curettage.

central area (Fig 4, A). After three years of the electrosurgery session, the scar became mildly dyschromic, showing alternate hypochromic and hyperchromic features (Fig 4, B).

Retrospectively the agent was identified as *S. brasiliensis* by a species-specific polymerase chain reaction method.³ The minimal inhibitory concentrations in the antifungal susceptibility test performed according to the Clinical & Laboratory Standards Institute M38-A2 document⁴ were as follows: itraconazole, 8 $\mu\text{g/ml}$; terbinafine, 0.5 $\mu\text{g/ml}$; posaconazole, 8 $\mu\text{g/ml}$; and amphotericin B, 4 $\mu\text{g/ml}$.

DISCUSSION

Sporotrichosis is considered as refractory to systemic treatment when there is no clinical improvement in the patient's condition even after 4 months of pharmacological treatment or in cases with stagnant

therapeutic response.^{1,2} Microbial culture performed in the laboratory is a fundamental requirement to confirm the diagnosis of sporotrichosis.

Itraconazole is the main drug used for the treatment of sporotrichosis because of its efficacy, safety, and dosage convenience. However, some strains of *Sporothrix brasiliensis* show a greater tolerance to this drug.^{1,5-7}

The causative species in this patient, *S. brasiliensis*, corroborates the molecular epidemiological studies conducted in this region, although it was not caused by zoonotic transmission. Anatomical and clinical-functional conditions, such as the location of lesions on the lower limb, human immunodeficiency virus infection, fibrotic process during treatment, drug interactions, and low intestinal absorption, may have contributed to the ineffectiveness of the treatment.

Terbinafine is a safe and effective alternative in cases where itraconazole is contraindicated.^{8,9} Potassium iodide can be used in some special cases,⁵ whereas amphotericin B is used to treat systemic sporotrichosis.^{1,8} This patient responded slowly to itraconazole treatment, except for one lesion. Based on our experience and the patient's response, it was decided to modify the treatment by using local adjuvant therapy (such as cryosurgery, thermotherapy, and electrosurgery). Interestingly, the patient isolate showed an antifungal resistance profile in vitro, with high minimal inhibitory concentrations for the tested antifungals. However, these tests were performed to analyze the resistance profile, after the outcome, and did not guide our conduct.

Cryosurgery and thermotherapy are used as adjuvant therapies because *Sporothrix* sp. is sensitive to extreme temperatures. Cryosurgery provides excellent results; however, it often requires multiple sessions and the patient may experience localized pain, swelling, blisters, and dyschromia.⁶ Thermotherapy is less invasive and well tolerated but time and patient dependent.

Electrosurgery is based on the transmission of high-frequency electrical current through the tissues followed by their thermal denaturation.⁷ It is a

simple, low-cost, and safe technique indicated for plaques and crusted lesions.^{1,6} It was proven to be effective in maintaining the function and aesthetics of the operated site.

We emphasize that adjuvant therapies that are used for the treatment of infectious diseases aim to reduce the amount, time, cost, and adverse effects of pharmacological treatment, therefore aiding in clinical cure.

Conflicts of interest

None disclosed.

REFERENCES

1. Orofino-Costa R, Macedo PM, Rodrigues AM, Bernardes-Engemann AR. Sporotrichosis: an update on epidemiology, etiopathogenesis, laboratory and clinical therapeutics. *An Bras Dermatol*. 2017;92(5):606-620.
2. Soeiro Sampaio FM, Sguissardi de Oliveira D, Saraiva Freitas DF, Francesconi do Valle AC. Electrosurgery as adjuvant therapy for cutaneous sporotrichosis. *Dermatol Surg*. 2020;46(1):140-142.
3. Rodrigues AM, de Hoog GS, de Camargo ZP. Molecular diagnosis of pathogenic *Sporothrix* species. *PLoS Negl Trop Dis*. 2015;9(12):e0004190.
4. Clinical and Laboratory Standards Institute. *Reference Method for Broth Dilution Antifungal Susceptibility Testing of Filamentous Fungi; Approved Standard*. CLSI document M38-A2. 2nd ed. Clinical and Laboratory Standards Institute; 2008:52p.
5. Macedo PM, Lopes-Bezerra LM, Bernardes-Engemann AR, Orofino-Costa R. New posology of potassium iodide for the treatment of cutaneous sporotrichosis: study of efficacy and safety in 102 patients. *J Eur Acad Dermatol Venereol*. 2015;29(4):719-724.
6. Fichman V, do Valle ACF, Freitas DFS, Sampaio FMS, Lyra MR, de Macedo PM, et al. Cryosurgery for the treatment of cutaneous sporotrichosis: experience with 199 cases. *Br J Dermatol*. 2019;180(6):1541-1542.
7. Taheri A, Mansoori P, Sandoval LF, Feldman SR, Pearce D, Williford PM. Electrosurgery: part II. Technology, applications, and safety of electrosurgical devices. *J Am Acad Dermatol*. 2014;70(4):607.e1-607.e12.
8. Almeida-Paes R, Oliveira M, Freitas D, do Valle ACF, Galhardo MC, Zancoppe-Oliveira R. Refractory sporotrichosis due to *Sporothrix brasiliensis* in humans appears to be unrelated to in vivo resistance. *Med Mycol*. 2017;55:507-517.
9. Francesconi G, Valle AC, Passos S, Reis R, Galhardo MC. Terbinafine (250 mg/day): an effective and safe treatment of cutaneous sporotrichosis. *J Eur Acad Dermatol Venereol*. 2009;23(11):1273-1276.