

1 SUBMITTED 30 MAR 22
2 REVISION REQ. 14 JUN 22; REVISION RECD. 16 JUN 22
3 ACCEPTED 21 JUL 22
4 **ONLINE-FIRST: AUGUST 2022**
5 **DOI: [https://doi.org/ 10.18295/squmj.8.2022.048](https://doi.org/10.18295/squmj.8.2022.048)**

6
7 **Long-standing Lymphocutaneous Sporotrichosis**
8 **Israel Perez-López and *Ricardo Ruiz-Villaverde**

9
10 *Department of Dermatology, Hospital Universitario San Cecilio, Inst Invest*
11 *Biosanitaria IBS, Granada, Spain*

12 **Corresponding Author's e-mail: ismenios@hotmail.com*
13

14 **Introduction**

15 A 36-year-old male, farmer, with no relevant personal or family medical history ,
16 attended our Dermatology outpatient clinic, complaining of a warty plaque on the back
17 of his right hand that involved the entire dorsal aspect of the third finger and that
18 presented two satellites lesions on the arm with a linear arrangement (Fig.1.).

19 Locoregional adenopathies were not noted. He recalled a previous traumatic history
20 performing his work 10 months earlier. Two weeks later the lesions began to develop
21 until they reached their current state.

22
23 Laboratory examinations included blood sample test (haemogram and basical
24 coagulation), general biochemistry (lipid profile, hepatic and renal function),
25 autoimmunity (autoantibodies, immunoglobulins, complements), thyrotropin and
26 thoracoabdominal CT was requested to evaluate systemic dissemination with normality
27 of all the tests mentioned. No skin biopsy was performed. Regarding the initial clinical
28 suspicion of sporotrichosis, we started treatment with Itraconazole 200mg/12h, which
29 had to be discontinued due to gastrointestinal discomfort 10 days later. The study by
30 PCR assay of *M. tuberculosis* complex and Non tuberculous *Mycobacterium* spp was
31 negative. However, culture showed positive results for *Sporothrix schenckii* in five days
32 (being reported two days later). One week later the patient began treatment with
33 liposomal Amphotericin B at a dose of 5 mg/kg/day for 3 days as an induction regimen

34 and then once monthly as maintenance treatment until resolution of the clinical course,
35 four months later with no skin lesions. There have been no recurrences of the lesion six
36 months after the end of treatment. Written informed consent of the patient for
37 publication purposes has been obtained.

38

39 **Comment**

40 Sporotrichosis is a subcutaneous mycosis caused by a dimorphic fungus of the genus
41 *Sporothrix*. Sporotrichosis can be observed in any part of the world, with some areas of
42 "hyperendemicity", being particularly frequent in tropical and subtropical countries (in
43 Peru the reported incidence is 1/1,000 cases/year, while in the United States it is 1- 2
44 cases per million) due to more occupational exposure.¹

45

46 Without predilection for age, sex or race, the occurrence of the disease depends on the
47 fungus being in the environment and traumatic inoculation into the skin . The male
48 predominance is believed to be due to greater exposure rather than greater
49 predisposition. Traumatic inoculation is the reason why the extremities (particularly
50 upper extremities) and bare parts are affected most often. Single or multiloculated
51 cutaneous forms, lymphocutaneous, as in the case at hand, and systemic (may
52 compromise lung, breasts, liver, kidney, eyes, heart, and genitalia) have been
53 described.² Culture in Sabouraud Dextrose Agar continues to be the gold standard, since
54 histological study may be less profitable and require histochemical techniques such as
55 PAS, Grocott or Gomori to identify fungal structures. In some reference centers, PCR
56 assay is used for its diagnosis, although the kits are not commercially available. The
57 differential diagnosis following a sporotrichoid eruption pattern is wide, encompassing
58 cutaneous tuberculosis, leishmaniasis, nocardiosis, chromoblastomycosis,
59 blastomycosis, paracoccidioidomycosis, and atypical mycobacteriosis.

60

61 The first line of treatment in the lymphocutaneous variety is oral itraconazole
62 100mg/day.³ Oral terbinafine has been used with moderate success and in more
63 resistant cases the use of oral saturated solution of potassium iodide (SSKI) has been
64 reported. This last modality lacks a standardized commercial formulation and its
65 metallic taste and uncertain mechanism of action have relegated it to a second or third
66 line choice. Liposomal amphotericin B may be a more effective option in
67 immunosuppressed patients or those with low therapeutic adherence.⁴ Among its side

68 effects are fever, headache, malaise, hypokaliemia, hypomagnesemia, cardio and
69 nephrotoxicity. The use of other less conventional treatments such as photodynamic
70 therapy has only been carried out in case series and with uneven results.⁵

71

72 **Author Contributions**

73 IPL and RRV were both involved in conceptualization, collection of data, writing and
74 final approval of the manuscript.

75

76 **References**

- 77 1. Sharma B, Sharma AK, Sharma U. Sporotrichosis: a Comprehensive Review on
78 Recent Drug-Based Therapeutics and Management. *Curr Dermatol Rep*. 2022
79 Mar 17;1-10. doi: 10.1007/s13671-022-00358-5.
- 80 2. Martínez-Herrera E, Arenas R, Hernández-Castro R, Frías-De-León MG,
81 Rodríguez-Cerdeira C. Uncommon Clinical Presentations of Sporotrichosis: A
82 Two-Case Report. *Pathogens*. 2021 Sep 27;10(10):1249. doi:
83 10.3390/pathogens10101249.
- 84 3. Poester VR, Basso RP, Stevens DA, Munhoz LS, de Souza Rabello VB,
85 Almeida-Paes R, Zancopé-Oliveira RM, Zanchi M, Benelli JL, Xavier MO.
86 Treatment of Human Sporotrichosis Caused by *Sporothrix brasiliensis*. *J Fungi*
87 (Basel). 2022 Jan 10;8(1):70. doi: 10.3390/jof8010070.
- 88 4. Belda W Jr, Domingues Passero LF, Stradioto Casolato AT. Lymphocutaneous
89 Sporotrichosis Refractory to First-Line Treatment. *Case Rep Dermatol Med*.
90 2021 Oct 6;2021:9453701. doi: 10.1155/2021/9453701.
- 91 5. Legabão BC, Fernandes JA, de Oliveira Barbosa GF, Bonfim-Mendonça PS,
92 Svidzinski TIE. The zoonosis sporotrichosis can be successfully treated by
93 photodynamic therapy: A scoping review. *Acta Trop*. 2022 Apr;228:106341.
94 doi: 10.1016/j.actatropica.2022.106341.

95



96

97 **Fig. 1:** Verrucous plaque on the back of the right hand that involved the entire dorsal
98 aspect of the third finger with two satellites lesions on the arm with a linear
99 arrangement.