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A case of disseminated sporotrichosis caused by *Sporothrix brasiliensis*

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ABSTRACT

This paper presents a case of disseminated sporotrichosis in a 13-year-old female, originating from a rural area in Minas Gerais state, Brazil. The patient was hospitalized in Santa Casa hospital of Belo Horizonte, with hyporexia, prostration, fever and disseminated ulcerative lesions, besides anemia, leucopenia and sepsis of probable cutaneous focus. The patient was admitted without proven immunosuppression. She was diagnosed with cutaneous-disseminated sporotrichosis. The drug therapy chosen was itraconazole during 12 months, leading to important clinical improvement and healing of cutaneous lesions.

1. Introduction

Sporotrichosis is a subcutaneous mycosis caused by dimorphic fungi of the genus *Sporothrix*, with global distribution [1–3] but mostly reported from Asia, South America and South Africa [4]. In humans, the infection classically occurs by the traumatic inoculation from contaminated sources, such as woods, thorns and splinters, and hence is known as "gardener's disease". The infection can also occur through scratches and bites of infected cats [5].

Cutaneous sporotrichosis is the most frequent form of the disease [6,7]. The pulmonary form may be confused with tuberculosis in radiological findings. The lesions may be confined to the site of inoculation or reach the lymphatic system, leading to the disseminated form [8]. Immunocompromised or alcoholic patients are the most susceptible to the visceral and osteoarticular forms [9].

The infectious species in the genus *Sporothrix* are *Sporothrix brasiliensis, S. schenckii, S. lurieri, S. globosa, S. mexicana* and *S. pallida* [10]. Of these, *S. brasiliensis* is restricted to Brazil and *S. globosa*, though distributed worldwide, has a higher incidence in East Asia [11].

Diagnosis can be made through direct microscopic examination, culture, histopathological examination, serology and intradermal reaction (sporotrichine test) [12]. Molecular diagnosis of the disease is done by partial sequencing of rDNA ITS or the partial calmodulin gene, showing an efficient, safe and fast identification [13,14].

Sporotrichosis rarely presents spontaneous remission. Therefore, adequate treatment is required [15]. According to the Infectious Diseases Society of America (IDSA), itraconazole is the indicated treatment for the lymphocutaneous and disseminated forms of the disease [16]. Alternative treatment of the cutaneous includes saturated potassium iodide [17]. Treatment with amphotericin B is indicated only for severe infections, but the the general health state of the patient should then be carefully assessed, because of high toxicity of the antifungal [6].

In this study, we present the case report of a disseminated sporotrichosis in an adolescent, in which the diagnosis was confirmed through positive culture as well as by sequencing ITS rDNA.

2. Case report

Female, 13 years old, originating from a rural area, was admitted in September of 2015 for hospitalization presenting hyporexia, prostration, febrile neutropeny, besides anemia and leucopenia. The patient presented ulcerated erythematopapular lesions without lymphadenopathy, with sepsis of probable cutaneous focus (Fig. 1A). Results from non-reactive PPD tests and serology for HIV, VDRL and leishmaniosis

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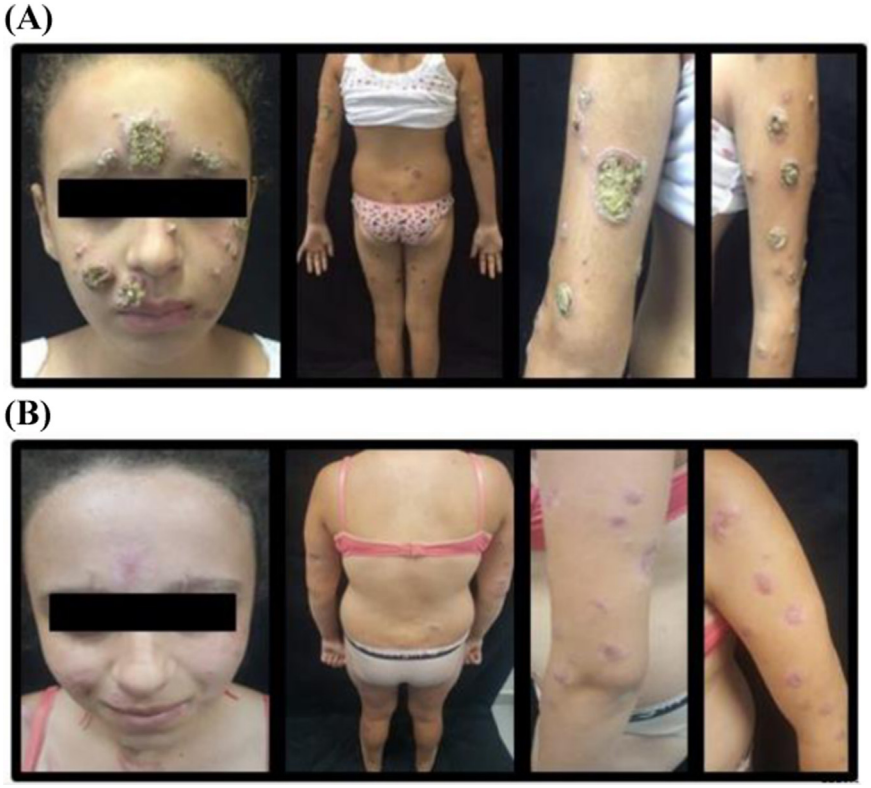

Fig. 1. (A) Clinical presentation of lesions spread throughout the body. (B) Clinical presentation of lesions after treatment.

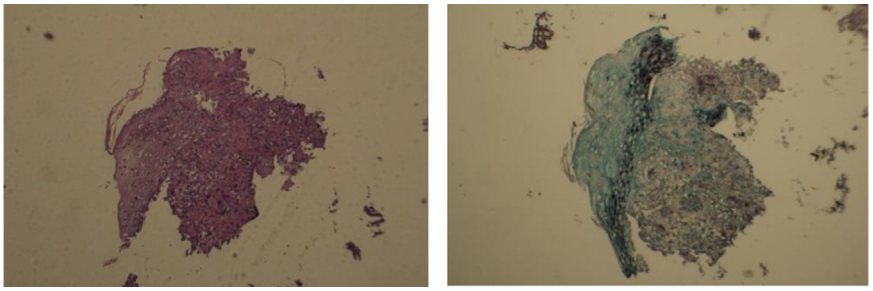

Fig. 2. Histopathological examination stained with Hematoxylin - Eosin (A) and stained with Silver (B), both of which have granulomatous dermal infiltrate.

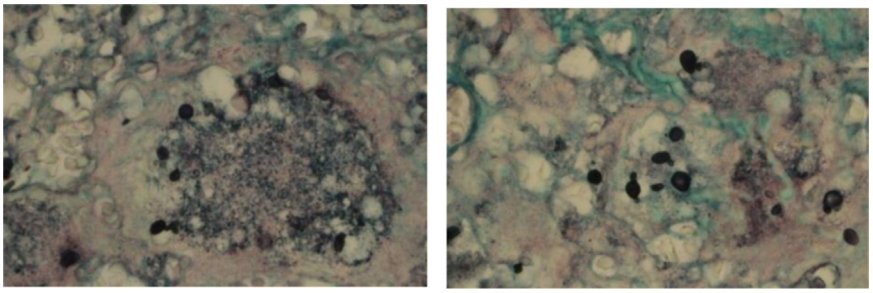

Fig. 3. Histopathological examination stained with Prat, both showing rounded refractive structures.

were all negative. Serological tests for hepatitis, hemoculture and prophylaxis for hemolytic anemia all showed negative results. Hepatic and renal functions presented no changes. The myelogram showed decreased megakaryocytic series and EDA a mild and non-specific chronic pangastritis. Tomography of the skull and sinuses as well as chest showed no changes. The biopsy showed dermal granulomatous infiltrate (Fig. 2), multinucleated giant cells and rounded refractive structures (Fig. 3). Yeast-like cells were observed inside giant cells (Fig. 4), and an entire arsenal of the immune-cellular response linked to the presence of fungus in host tissues was noted. Judging from clinical evaluation of hematology / oncology, the possibility of hematological neoplasia was ruled out. The changes in the myelogram were compatible with a medullary reaction state due to systemic infection.

Culture was performed on Sabouraud's Agar containing 0.05% chloramphenicol, presenting growth of a creamy, brownish beige colony, typical of *Sporothrix* spp. Microculture showed thin septate hyphae containing conidia in a dense arrangement at its extremities (data not show). The culture was deposited at "Microbial Collections of

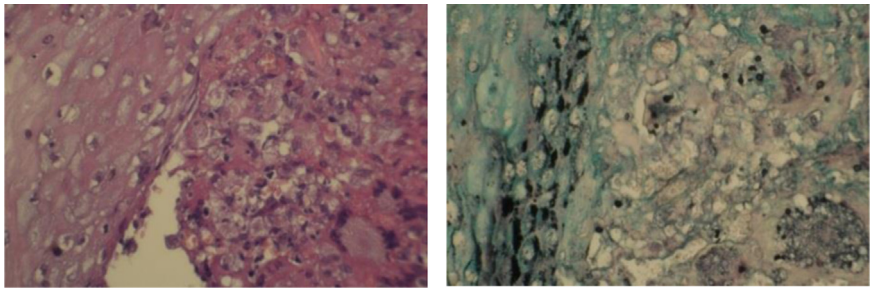

Fig. 4. Histopathological examination stained with Hematoxylin - Eosin (A) and stained with Silver (B), both presenting yeast-like cells inside the giant cells.

Paraná, Network - TAXon line", with accession number CMRP1416 on day 30. The fungal culture was submitted to DNA extraction, with subsequent sequencing of the ribosomal region ITS1-ITS2 of the rDNA [10,18], GenBank accession number: KX247390. A phylogenetic tree was constructed using ITS sequences), with comparison with reference strains of the genus *Sporothrix*, confirming species identification as *Sporothrix brasiliensis*.

The patient was consequently diagnosed with cutaneous-disseminated sporotrichosis, without involvement of internal organs. On day 40 after admission, treatment with itraconazole 400 mg/day was started. The dose of itraconazole was adjusted to 200 mg/day on day 60 and patient was discharged on day 120 after admission for home treatment and follow up with dermatology. Treatment continued during 12 months, presenting important clinical improvement, involution and healing of cutaneous lesions (Fig. 1B).

3. Discussion

Sporotrichosis is caused by members of the dimorphic genus *Sporothrix*, with appearance of lesions in cutaneous and subcutaneous tissues, and may affect lymph nodes that eventually rupture to the skin [18]. The disease may vary from subacute to chronic and its clinical manifestation and severity are related to the host immunity, fungal load and *Sporothrix* genotype [19].

The patient reported in the present case had widespread cutaneous lesions that probably had disseminated via the hematogenic pathway but without any involvement of internal organs. Patient had no proven immune disorder. This matched with the observation that, as soon as the therapeutic regimen was started, the patient responded promptly, leading to decrease of the lesions within a week. At the end of the treatment she had remission of all lesions and remained healthy during follow-up (Fig. 1B).

The present case demonstrates that the specialist should be aware of this exuberant form of sporotrichosis, despite the fact that the lymphocutaneous presentation is the most commonly observed form in immunocompetent patients [6]. The professional should also note that some *Sporothrix* species are more virulent, as among which is particularly *S. brasiliensis*, the causative agent identified in this case [20].

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Conflict of interest

The authors have no conflicts of interest to declare and confirm that each one has made substantial contributions to the information or materials submitted for publication.

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