Familial occurrence of zoonotic sporotrichosis

Ocorrência familiar de esporotricose zoonótica

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Abstract: Sporotrichosis is a subacute or chronic mycosis caused by the dimorphic fungus Sporothrix schenckii which is endemic in Brazil and is transmitted primarily through traumatic inoculation of its causative agent into the skin. The zoonotic transmission, especially from infected cats, has been demonstrated in several reports and case series. We present simultaneous occurrence of the disease in three members of the same family by scratches from an infected domestic cat. Two patients developed the lymphocutaneous form and one only developed the fixed cutaneous form. Two patients were successfully treated with saturated solution of potassium iodide; however, the third case reported side effects and had his therapy substituted for itraconazole, with resolution of his lesions.
Keywords: Cats; Disease transmission, infectious; Potassium iodide; Sporotrichosis

Resumo: A esporotricose é micose subaguda ou crônica, causada pelo fungo dimórfico Sporothrix schenckii, endêmica no Brasil e transmitida principalmente através da inoculação traumática de seu agente causal na pele. A transmissão zoonótica, especialmente por gatos infectados, tem sido demonstrada em diversos relatos e séries de casos. Nós descrevemos a ocorrência simultânea da doença em três membros de uma mesma família através da arranhadura por gato doméstico infectado. Dois pacientes desenvolveram a forma cutânea-linfática e apenas um desenvolveu a forma cutânea fixa. Dois pacientes foram tratados com sucesso, com solução saturada de iodeto de potássio; entretanto, o terceiro caso apresentou efeitos colaterais e teve seu tratamento substituído por itraconazol, com resolução de suas lesões.
Palavras-chave: Esporotricose; Gatos; Iodeto de potássio; Transmissão de doença infecciosa

INTRODUCTION

Sporotrichosis is a cutaneous, subcutaneous or systemic mycosis caused by the dimorphic fungus *Sporothrix schenckii.* The main clinical presentations of the disease are the fixed cutaneous and lymphocutaneous forms. The disease is widely distributed throughout the world and it is considered endemic in South America. The classical transmission is via traumatic inoculation of the fungus into the skin. The disease is considered by some authors as occupational. [1] The number of cases of zoonotic transmission has been increasing significantly, with a recent report of an epidemic outbreak with documented zoonotic transmission by infected cats in Rio de Janeiro, south east of Brazil. [2]

In this paper we report the occurrence of simultaneous cases of sporotrichosis in three members of the same family, where the source of infection was a domestic cat.

CASES REPORT

Case 1: White female, 35 years of age, single, physiotherapist, born and resident in DF (urban area) who in 2009 presented with an ulcer on the 5th left finger and lateral border of the left arm, erythematous

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nodules on the same arm following the lymphatic channels, which were tender to palpation, as well as grouped papules and pustules on the right malar region and upper lip, of a month´s duration (Figure 1A). She reported scratches by a domestic cat, which had been put down 2 months before because of an undiagnosed cutaneous disease. *Sporothrix schenckii* was isolated after cultivation. The histopathologic examination of the biopsy from the left hand showed an area of ulceration with fibrinous leucocytes exsudate, heavy lymphohystiocitic infiltrate, as well as giant cell granulomas and foci of exsudate on the dermis. PAS staining showed few round yeast forms with pale center and darker periphery and isolated, focal buds (Figure 1B). The patient received saturated solution of potassium iodide (dose: 3 g/day), with complete resolution of the lesions in seven months (Figure 1C).

Case 2: 60 years old female, married, housewife, mother of patient 1, who lived in the same house. During consultation with the dermatologist she presented with an infiltrated, erythematous plaque on the distal third of the right arm, one month after being scratched by a domestic cat (Figure 2A). *Sporothrix schenckii* was isolated from culture. The histopathologic examination showed hyperkeratosis, spongiosis, pseudoepitheliomatous hyperplasia and exocytose of leucocytes. The papillary dermis showed mixed inflammatory infiltration with foreign body giant cells and microabcesses. Staining for fungus with PAS and Grocott were negative. The patient received saturated solution of potassium iodide (dose: 3 g/day) with healing of the lesions in three months.

Figure 1: Case 1 - Papules and pustules located above the lip and on the malar area;

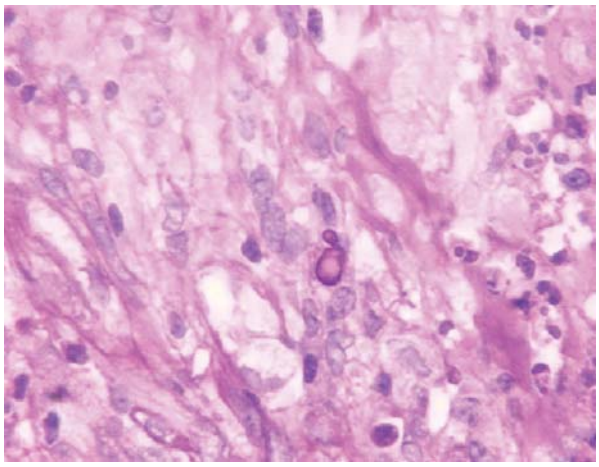

Figure 2: Case 1 - Presence de round yeasts with a pale centre and reinforced coloration on the periphery showing budding, surrounded by lymphohystiocitic infiltrate (PAS with diathesis, 40x);

Figure 3: Case 1: Healing after seven months´ treatment with saturated solution of potassium iodide;

Case 3: 74 years old male, retired, father of patient 1, who lived in the same house of patients 1 and 2. He presented with an ulcer on the 5th finger of the right hand, and two erythematous, tender nodules, following the lymphatic channels on the right arm (Figure 2B). *Sporothrix schenckii* was isolated from culture and the histopathologic examination showed acanthosis, focal parakeratosis, pseudoepitheliomatous hyperplasia and exocytose of the lymphocytes. There was a diffuse monocytic inflammatory infiltrate on the dermis, with epithelioid cells and rare multinucleated giant cells. Staining for acid-fast bacterium and fungus were negative. The patient was treated with saturated solution of potassium iodide (dose: 3g/day) for 4 months with partial improvement. Due to gastric and intestinal intolerance and

Figure 4: Case 2 – Mother of the index case presenting with an erythematous plaque on the right forearm;

Figure 5: Case 3 – Father of the index case presenting with nodular, ulcerated lesions along the lymphatic channels on the forearm;

insomnia the treatment was changed to itraconazole (dose: 100mg/day) with complete resolution of the lesions after 2 months of treatment.

DISCUSSION

Sporotrichosis is a subacute or chronic mycosis, caused by *Sporothrix schenckii*, a dimorphic fungus usually found on the soil and in decomposing organic material. Sporotrichosis was originally described by Schenck in 1898. [3] The first case of the disease in Brazil was reported by Lutz and Splendore in 1907. [4]

The disease is widely distributed throughout the world and it is considered the most common subcutaneous mycosis in South America. A great number of cases and some indication of endemicity have been reported in countries like Mexico, Japan, Brazil, Peru and Colombia. [2,4] Rosa and collaborators classified the south region of Brazil as an endemic area for sporotrichosis. [5]

The classic transmission of the disease is via traumatic inoculation of the fungus into the skin through contact with contaminated material. The disease is sometimes classified as an occupational hazard and professionals like florists, horticulturists, gardeners and miners have a higher risk of getting infected. [1, 6] Other forms of contamination include inhalation of the fungus and zoonotic transmission through scratches or bites from domestic animals, mostly cats.

There are various clinical presentations and they depend on facts like the host's immunological status, the size of the infective inoculum and the virulence of the fungus, although the fixed cutaneous and lymphocutaneous forms are the most commonly described. [1] The disseminated cutaneous presentation of sporotrichosis has been observed mainly in immunocompromised patients, especially HIV positive ones. Extra cutaneous clinical presentations such as pulmonary, osteoarticular, ocular, and meningitis are rare. [7]

The diagnosis is based on the clinical history and the isolation of the fungus via culture in medias such as Agar Sabouraud-dextrose or Agar dextrose-potato, incubated at 25°C. The growth of the colonies can be seen in 30 days. [1] The media BHI Agar can be used to identify the thermal dimorphism. [2] The histopathological examination is not specific; however it is a useful tool to diagnosis by showing the pseudoepitheliomatous hyperplasia and the granulomatous reaction, which can harbor microabscesses within. With the use of special stainings like PAS, yeast aspects of *S. schenckii* like cigar-shaped structures and budding yeasts, as well as extracellular asteroid bodies, can be identified. [7, 8] Serologic testes can be useful on the diagnosis of sporotrichosis, especially in atypical extra-cutaneous presentations. [4]

In the cases presented here the likely source of infection was the infected domestic animal. However, despite the notification to the public organs by the patients, the cat was incinerated without investigation and confirmation of the diagnosis. This report corroborates recent data from the literature that alert to the increasing number of zoonotic transmission, like the ones observed by Barros and collaborators in an epidemic in the city of Rio de Janeiro. [2] In his paper 178 cases of the disease in humans were diagnosed from 1998 to 2001, from which 90.7% reported familial or professional contact with infected cats and 64.5% reported history of traumatic injury preceding the beginning of the symptoms.

In the present cases, despite a common source of infection two patients developed the cutaneous-lymphatic form of the disease and one developed only the fixed cutaneous form, thus emphasizing the evidence that facts like the size of the inoculum and the host's immune response can influence the clinical presentation.

The histopathological examination in case 1 identified, through PAS staining, the yeasts with buds.

This finding has been described in the literature as rare. However, Barros and collaborators identified the presence of fungal elements in 28.8% of the histopathological examinations and formulated the hypothesis that this high percentage could be related to the zoonotic transmission, what can be corroborated by the exuberance of fungal elements found in cutaneous lesions of infected cats. [2, 8, 9]

There are many treatment modalities for sporotrichosis, from exposure to heat to the use of saturated solution of potassium iodide and antifungal like itraconazole, but according to the medical literature the success rates are variable.

The saturated solution of potassium iodide has been used since 1900, with success rates varying from 80 to 100%. [10] A recent Cochrane analysis preformed by Xue and collaborators evidenced the lack of randomized and placebo-controlled studies in this field and the authors admitted that the available evidence were insufficient to confirm the efficacy of the potassium iodide on the treatment of sporotrichosis. [11] The guidelines for the management of sporotrichosis published by Kauffman and collaborators in 2007 suggest the itraconazole at the dose of 200mg a day as the treatment of choice for the fixed cutaneous and cutaneous-lymphatic forms of the disease. However, this is on the same level of recommendation (A-II) for the saturated solution of potassium iodide. [10]

The choice of the saturated solution of potassium iodide in these cases was based on reports from the literature, the experience of the service and the low cost. As a confirmation of the efficacy of this medication two patients had complete clinical resolution of the lesions. This fact could not be observed with patient 3 due to side effects, and his treatment was changed to itraconazole.

Sporotrichosis remains an endemic mycosis in Brazil, however the real incidence of this disease in the country remains unknown. We reported the uncommon and simultaneous occurrence of sporotrichosis in three members of the same family, living in the same house, transmitted by an infected domestic animal. ❑

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