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A rare case of oral sporotrichosis: painful nodule in the buccal mucosa

Um caso raro de esporotricose oral: nódulo doloroso na mucosa jugal

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RESUMO

A esporotricose é uma infecção fúngica comum que apresenta várias manifestações clínicas, incluindo formas cutâneas, mucosas, osteoarticulares, sistêmicas e imunorreativas. Sua ocorrência na cavidade oral é incomum, sendo mais frequentemente observada em pacientes imunocomprometidos e manifestando-se como lesões ulcerativas. No presente caso, uma paciente de 62 anos apresentou queixas de dor na mucosa bucal. No exame extraoral, foram notados nódulos com uma área ulcerada e crostosa no punho e antebraço esquerdo, além de uma úlcera localizada no lado medial do calcanhar direito. O exame intraoral revelou um nódulo doloroso com coloração normal na mucosa bucal esquerda. Inicialmente, foi realizada uma biópsia incisional, que revelou um infiltrado inflamatório difuso com áreas de necrose. Posteriormente, foram realizadas coloração com Ácido Periódico de Schiff (PAS) e cultura celular, que revelaram estruturas consistentes com *Sporothrix spp*. Com base nesses achados, foi estabelecido o diagnóstico de esporotricose e escolhida a abordagem terapêutica com itraconazol 100 mg (1 comprimido a cada 12 horas) por 6 meses. A paciente apresentou resolução completa das lesões.

Palavras-chave: Esporotricose. Manifestações Bucais. Diagnóstico Bucal. Micoses. Relatos de Casos.

ABSTRACT

Sporotrichosis is a common fungal infection that presents with various clinical manifestations, including cutaneous, mucosal, osteoarticular, systemic and immunoreactives. Its occurrence in the oral cavity is uncommon, more frequently observed in immunocompromised patients, and manifests as ulcerative lesions. In the present case, a 62-year-old patient presented with complaints of pain in the buccal mucosa. Upon extraoral examination, nodules with an ulcerated and crusted area were noted on the left wrist and forearm, along with an ulcer located on the medial side of the right heel. Intraoral examination revealed a painful nodule with normal coloration on the left buccal mucosa. Initially, an incisional biopsy was performed, revealing a diffuse inflammatory infiltrate with areas of necrosis. Subsequently, Periodic Acid Shiff (PAS) staining and cell culture were performed, revealing structures consistent with Sporothrix spp. Based on these findings, the diagnosis of sporotrichosis was established, and itraconazole 100 mg (1 tablet every 12 hours) for 6 months was chosen as the therapeutic approach. The patient exhibited complete resolution of the lesions.

Keywords: Sporotrichosis. Oral manifestations. Oral diagnosis. Mycoses. Case Reports.

1. INTRODUCTION

Sporotrichosis is an infection caused by a dimorphic fungus of the gender Sporothrix spp. which has a global distribution.[1] The species already isolated and commonly known are S. brasiliensis, S. schenckii, S. globosa and S. luriei, which can be transmitted through contact with contaminated soil or zoonotically by domestic animals, especially cats.[2] The first transmission route is considered classical, as it refers to sapronosis and is associated with outdoor workers such as gardeners and farmers. The second transmission route, through animals, has been gaining attention due to the increasing number of domestic animals and the high zoonotic potential that directly influence the severity of the disease. This exposure results from traumatic inoculation, through scratches and bites or through contact with exudate from skin lesions of the animal.[3]

The immunological condition of the host may have a greater impact on the clinical manifestation.[2] For example, in the United States of America, there were about 1,471 hospitalizations due to sporotrichosis infection in patients who were seropositive, had immune-mediated inflammatory diseases or in patients with chronic obstructive pulmonary disease. In the literature, there are few manifestations of sporotrichosis lesions in the oral cavity. Interestingly, multiple papules and ulcerations are more common and immunocompromised patients are more affected.[4]

The occurrence of a sporotrichosis affecting oral tissues is very uncommon, although it may be the first manifestation of the infection.[5] In this paper, we bring a rare clinical presentation of this disease in the oral cavity, focusing on the most important diagnostic features in this interesting discussion.

2. MATERIALS AND METHODS

This work is a descriptive case report aimed at detailing the diagnostic and treatment process of a patient with sporotrichosis. The research was conducted with the appropriate approval from the Research Ethics Committee of the Lauro Wanderley University Hospital, affiliated with the Federal University of Paraíba (6.811.016).

3. CASE REPORT

A 62-year-old woman with no underlying medical conditions was referred to our service due to complaints of a painful lesion in the cheek region. During anamnesis, the patient reported that she had been scratched by a domestic cat about 40 days ago, and soon after, lesions appeared on her skin (hand and foot) and in her mouth. Extraoral examination revealed reddish nodules with an ulcerated and crusted area on the left wrist and forearm region, and an ulcer located on the medial side of the heel of the right foot (Figure 1A). In the head and neck region, an edema was observed in the left cheek region (Figure 1B). On intraoral examination, a palpable, symptomatic, normochromic nodule on the left cheek was noted (Figure 1C).

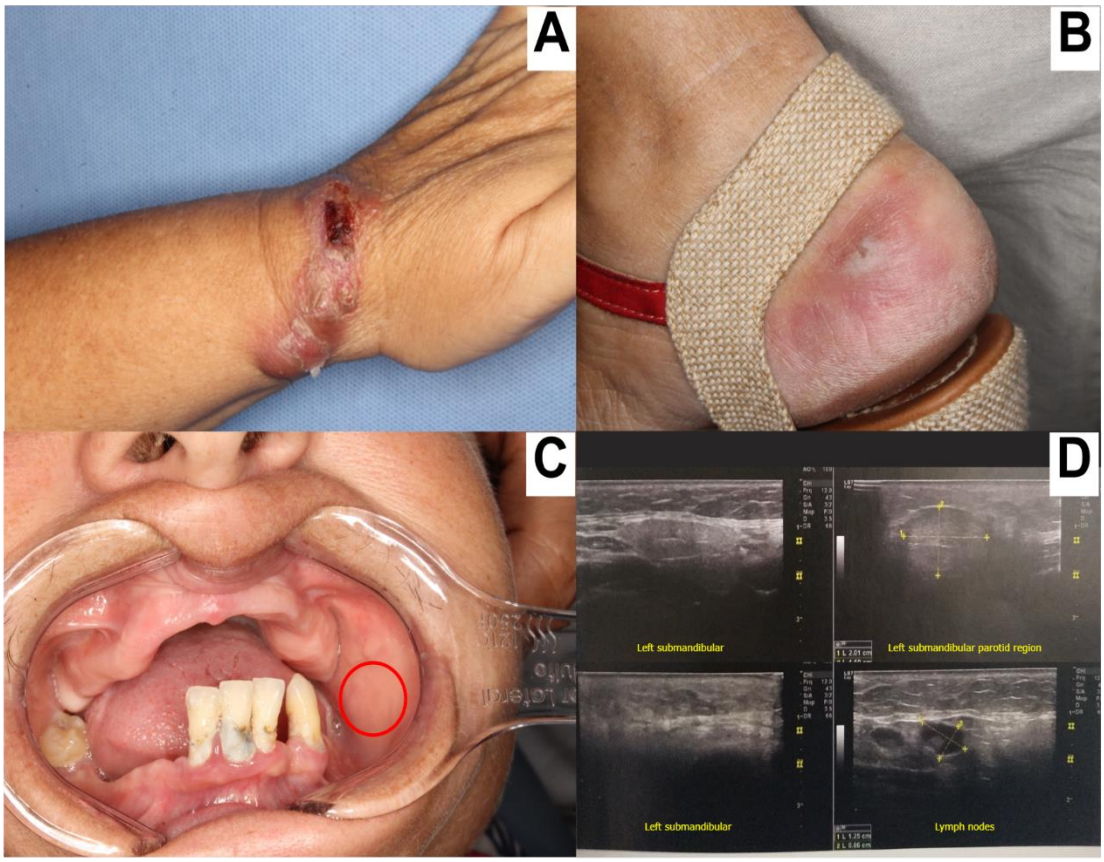

Figura 1. A) Erythema nodosum on left wrist and forearm region; B) An ulcer was observed on the right foot; C) Intraoral examination evidenced a palpable and normochromic nodule D) Ultrasound revealed a nodule in the left pre-parotid region, associated with edema, and enlarged lymph nodes.

Initially, an incisional biopsy of the nodule detected on the left cheek, along with an ultrasound, was requested. The parotid gland ultrasound revealed a superficial nodular formation in the left pre-parotid region, associated with locoregional edema, and enlarged

lymph nodes in the left cervical chains (Figure 1D). Histopathology showed a diffuse inflammatory infiltrate with some foci of tissue necrosis (Figure 2A).

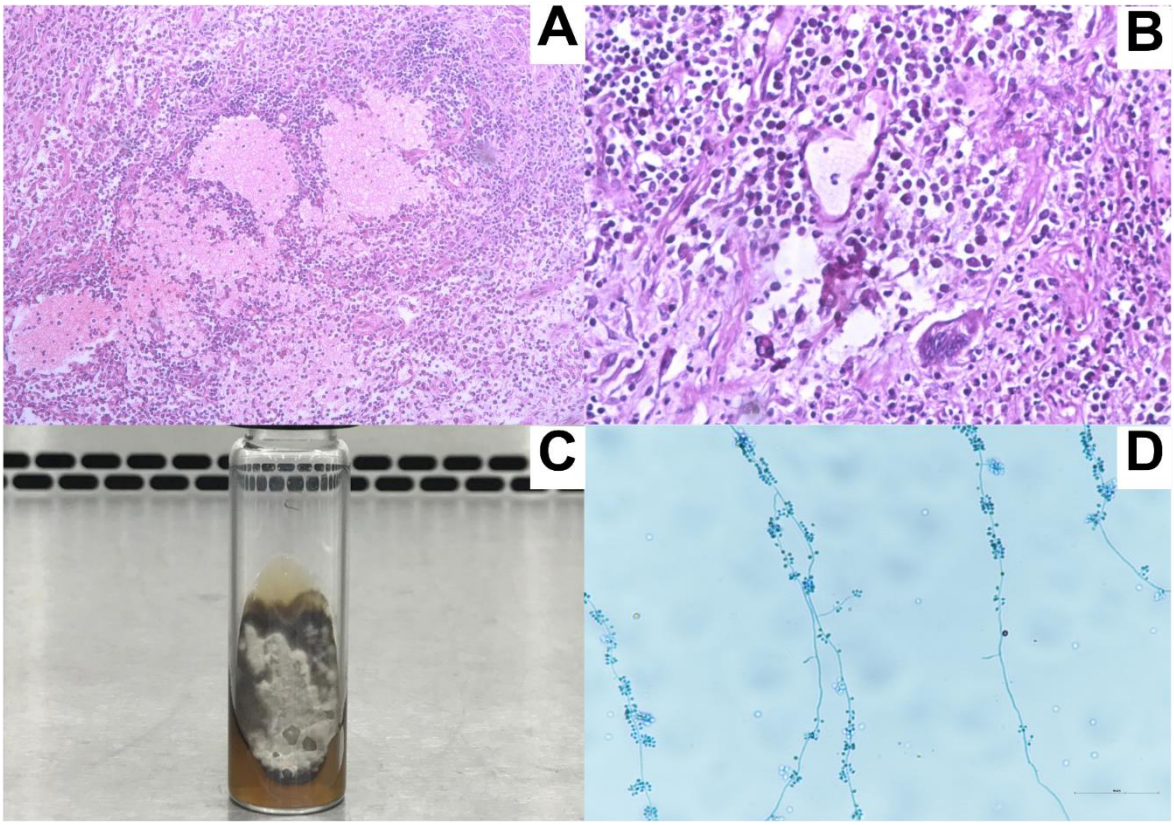

Figure 2. A) Incisional biopsy revealed a diffuse inflammatory infiltrate with tissue necrosis (Hematoxilin and eosin, 40x); B) Free yeasts, rounded and elongated, yeasts in the cytoplasm of macrophages and in buddings were observed through Periodic Acid Schiff (PAS) staining; C-D) After imprint technique, thin, hyaline, septate and branched hyphae containing denticles with conidia in flower-like arrangements were seen (cotton blue lactophenol).

After that, Sporothrix spp. was identified throughout a Periodic Acid Schiff (PAS) staining (Figure 2B). There, free yeasts, rounded and elongated, yeasts in the cytoplasm of macrophages and in buddings were observed. Afterwards, fungal culture was performed using the imprint technique on sabouraud agar (10 days at 25°C), where it was possible to observe the presence of thin, hyaline, septate and branched hyphae containing denticles with conidia in flower-like arrangements, stained with cotton blue lactophenol (Figure 2C-D). Based on this, the diagnosis of sporotrichosis was established, and the chosen therapeutic course was itraconazole 100 mg (1 tablet, every 12 hrs) for 06 months. The patient showed a complete resolution of the lesions after 3 months of treatment.

4. DISCUSSION

Considering the clinical presentation and location of the single submucosal nodular lesion, the diagnostic hypotheses were mesenchymal, glandular, and dermal, besides being a manifestation of infectious diseases. Based on a rapid growth, and a cat scratch history associated, the most likely diagnosis was an infectious lesion.

First, among the neoplasms of mesenchymal origin, lipoma was the main lesion considered.[6] This lesion presents as single, asymptomatic, slow-growing, well-circumscribed submucosal nodules. It may also exhibit normochromic coloration in deeper lesions and may be confused with other neoplasms.[7–8] Lipomas are more common in middle-aged individuals, with a slight female predilection.[8] Although the clinical features are suggestive of this lesion, the rapid appearance of the nodule discourages its diagnosis. Furthermore, solitary fibrous tumor was considered which has an intermediate biological behavior and may appear as an exophytic nodule (25.3%) or as a submucosal swelling (24.7%), with a high prevalence (44.8%) in the cheek region.

Salivary gland neoplasms represent about 1% to 4% of neoplasms. Pleomorphic adenoma is the most common salivary gland disease. This neoplasm occurs mainly in young and middle-aged (30 to 60 years old) females. In the minor salivary glands, the most common sites of involvement are the palate, upper lips, and cheek. Clinically, they are usually painless, slow growing, and firm on palpation. Nevertheless, like lipoma, the rapid onset disfavors its diagnosis. On the other hand, due to this rapid growth, a malignant salivary gland neoplasm can be considered like a mucoepidermoid carcinoma. It appears more in the parotid region, but intraoral cases are located on the palate or cheek and may manifest as a superficial submucosal nodule, which is generally bluish-red and is associated with pain, discomfort, or other symptoms.[10]

Regarding neoplastic processes of dermal origin, we consider pilomatrixoma as a diagnostic hypothesis. This disease is considered rare, it originates from the hair follicle and its occurrence in women is more frequently reported.[11] The most common clinical presentation is a firm subcutaneous nodule that in most cases is asymptomatic. Computed tomography and ultrasonography may help in the identification of the lesion and its relationship with soft tissues.

Finally, we can consider the infectious diseases that manifest themselves through lesions in the mouth. Among them, tuberculosis, and sporotrichosis were considered. Despite being a disease that has declined considerably compared to past centuries, Brazil

remains among the 30 countries with a high burden for tuberculosis (TB).[12] Tuberculosis could be classified as primary or secondary tuberculosis (ST). In oral mucosa, ST manifests as persistent ulcers differing from our case. However, if we consider the nodule as a lymph node manifestation, we may have a deeper lymphadenopathy tuberculosis in the cheek. Thus, we could keep TB as the diagnostic hypothesis.

Another lesion to be considered was sporotrichosis which is a fungal disease caused by the fungus *Sporothrix spp.* that also affects patients in tropical/subtropical regions. Usually in the multifocal or disseminated presentations, there are lesions in the oral mucosa that are characterized as non-specific ulcerations, occurring more frequently in immunocompromised patients.[4] In addition, given its deeper location within the cheek, this lesion may represent an incipient stage of lymphocutaneous manifestation. The contextual information regarding scratching by a cat significantly heightens the likelihood of the diagnosis in the current case.

Sporotrichosis can be transmitted through contact with contaminated soil or zoonotically by domestic animals, especially cats. The first case of transmission of sporotrichosis through cats was reported in 1952 and given the different clinical manifestations, sporotrichosis can be classified into cutaneous, mucosal, osteoarticular, systemic, immunoreactive and mixed localized.[13] The clinical forms of sporotrichosis depend on the size of the infectious inoculum, fungal virulence, depth of inoculation and immunological status of the host. The dermatological manifestations of sporotrichosis infection include nodular, papular, plaque or follicular lesions. Regarding extra cutaneous manifestations, the mucous membranes (nose and oropharynx), the central nervous system and the lung may be affected.[5]

Furthermore, it is important to reinforce that the immunological condition of the host may have a direct influence on the clinical manifestation of sporotrichosis.[2] Here, the manifestation was a submucosal nodule, and the patient was not immunosuppressed. Thus, we bring an uncommon manifestation that may delay the diagnosis of this disease.

The diagnosis of sporotrichosis is based on culture detection, and clinicopathological correlation.[2] Fine needle aspiration may be useful in the presence of nodules, although it is occasionally unspecific. Another procedure indicated is the biopsy that must be performed.[13] The isolation and identification of the fungus by culture is considered the standard method for the definitive diagnosis of sporotrichosis, from the visualization of blackened or yellowish-

beige colonies, according to the temperature.[14] Here, we performed a biopsy of the nodule and fungal culture in which *Sporothrix spp*. was identified.

The management of the patient depends on the identification of the clinical form and diagnosis of sporotrichosis. After identification, some drugs can be used in the treatment. Itraconazole is the first line of treatment for sporotrichosis, but other drugs also have antifungal activity such as potassium iodide, terbinafine and amphotericin B, the latter being used in systemic, pulmonary and osteoarticular forms.[15] In any case, it is important to understand that the disease has been gaining more and more attention worldwide. Here, the treatment of the present report was carried out with itraconazole for six months, obtaining a successful treatment without recurrence.

5. CONCLUSION

In view of what has been discussed, sporotrichosis is a common zoonosis in Brazil and is usually associated with immunosuppressed patients. This disease may affect the oral cavity generally through an ulceration, but a rapidly growing submucosal nodule may be a manifestation of this lesion. The diagnosis of this condition is challenging and biopsy in association with special stains (PAS), cell culture and medical history are essential. Moreover, the proposed therapy has proved to be effective due to the complete remission of the lesions.

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