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Case report

Ocular sporotrichosis: A frequently misdiagnosed cause of granulomatous conjunctivitis in epidemic areas

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ABSTRACT

Purpose: Sporotrichosis is a subcutaneous mycosis caused by *Sporothrix* sp., a dimorphic fungus. Although the cutaneous form is the most frequent form, the ocular presentation has been increasingly diagnosed in epidemic areas. We describe three cases of ocular sporotrichosis with the involvement of the ocular adnexa due to autoinoculation without trauma with successful antifungal treatment.
Observations: Patient 1: A 68-year-old woman presented with granulomatous conjunctivitis of the right eye with an ulcerated nodule on the right temporal region for 5 months. Patient 2: A 46-year-old woman with conjunctival hyperemia of the left eye with associated periorbital edema and erythema for the past 4 months was referred to the Dermatology Department due to an ulcerated nodule on the left malar region. Patient 3: A 14-year-old boy presented to the emergency department with inferior palpebral edema with a 5-day evolution. Specimens were obtained from the lesions of the three patients, and the cultures were positive for *Sporothrix* sp. The three cases were diagnosed as ocular sporotrichosis and were successfully treated with itraconazole (200–400 mg/d). Two of the three patients developed sequelae such as conjunctival fibrosis and symblepharon.
Conclusions and importance: We emphasize the importance of the ophthalmologist being familiar with the diagnosis and management of this rare and frequently misdiagnosed form of sporotrichosis.

1. Introduction

Sporotrichosis is a subcutaneous mycosis caused by dimorphic fungi of the *Sporothrix schenckii* complex.[1] Although the cutaneous form is more frequent in Rio de Janeiro and other endemic areas, the ocular presentation has been increasingly diagnosed.[2] The similarity with other clinical conjunctivitis may lead to a delay in treatment initiation, increasing the risk of sequelae in the eye and extending the time needed for healing. Thus, the knowledge of this form of sporotrichosis is paramount for early diagnosis, and more effective therapy and clinical evolution.

2. Findings

2.1. Case 1

A 68-year-old woman presented 5 months previously with conjunctival hyperemia associated with granulomatous conjunctivitis in the right eye with no history of trauma. The condition was suggestive of bacterial conjunctivitis, and she was treated with antibiotic eye drops. The patient showed no improvement, and subsequently developed a nodular lesion on the right temporal region, for which she sought treatment from the Dermatology clinic. The dermatologic and ophthalmologic examinations showed granulomatous conjunctivitis involving the inferior tarsal and bulbar conjunctiva of the right eye (Fig. 1) associated with exudative ulcerated nodule on the ipsilateral temporal region (Fig. 2). The exudate culture of the skin lesion was positive for *Sporothrix* sp. The

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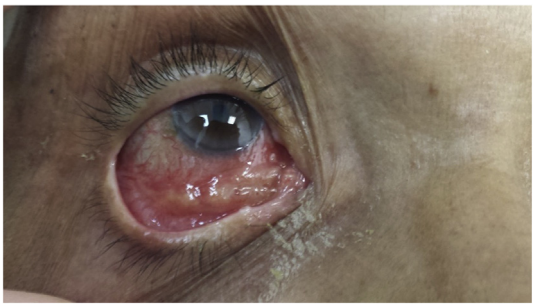

Fig. 1. Patient 1 - Granulomatous conjunctivitis of the right eye.

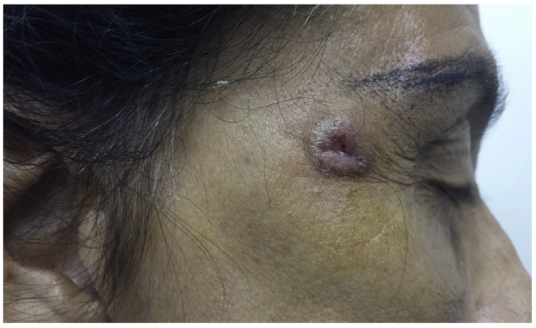

Fig. 2. Patient 1 - Nodule with central exulceration in the right temporal region.

patient was treated with itraconazole 200 mg/day over 9 months, achieving a clinical cure, but with fibrosis of the inferior tarsal and bulbar conjunctiva of the same eye (Fig. 3).

2.2. Case 2

A 46-year-old woman with conjunctival hyperemia associated with infiltration and periocular edema on the left side for 30 days was referred to the Dermatology clinic after a nodular lesion appeared on the ipsilateral malar region (Fig. 4). Dermatologic examination showed enlarged lymph nodes on the preauricular and cervical regions. The patient denied the occurrence of local trauma.

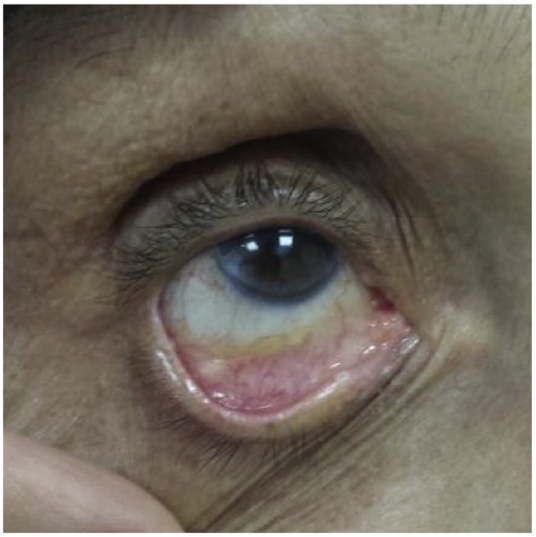

Fig. 3. Patient 1 - Fibrosis of tarsal and inferior bulbar conjunctiva of the right eye after treatment.

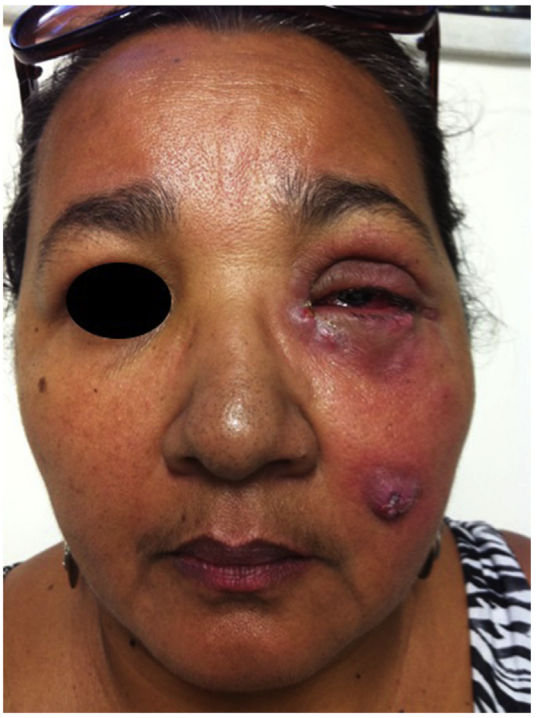

Fig. 4. Patient 2 - Conjunctival hyperemia associated with infiltration and periocular edema. Ulcerated nodules are noted in the ipsilateral malar region.

The swab culture of the conjunctival lesion was positive for *Sporothrix* sp. The patient was treated for 2 months with itraconazole 200 mg/day; however, it was necessary to increase the dose to 400 mg/day for an additional 6 months due to clinical worsening. As sequelae, she had fibrosis and symblepharon on the superior conjunctiva of the affected eye (Fig. 5).

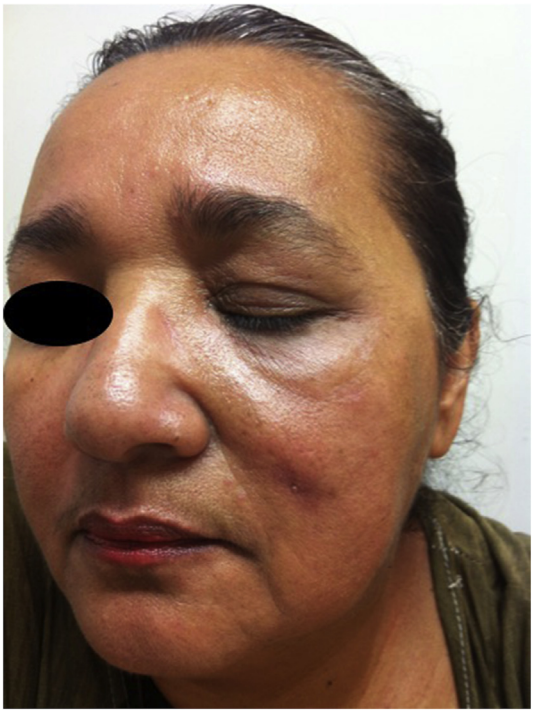

Fig. 5. Patient 2 - Eight months after treatment.

2.3. Case 3

A 14-year-old boy presented to the Emergency Department complaining of inferior right eyelid edema with a 5-day evolution, with no history of trauma of the conjunctiva. Physical examination showed granulomatous conjunctivitis affecting the lower tarsal conjunctiva of the right eye and enlarged lymph nodes on the preauricular and submandibular ipsilateral regions (Fig. 6). A swab of the conjunctival lesion was collected, and the culture was positive for *Sporothrix* sp. Treatment with itraconazole 200 mg/day was promptly started, leading to the disappearance of the lesions within 15 days (Fig. 7).

3. Discussion

Ocular involvement of sporotrichosis can occur in two ways: intraocularly by hematogenous dissemination, or by involvement of the ocular adnexa due to (self-)inoculation or trauma.[3]

Regarding the intraocular forms, four types have been described: granulomatous uveitis, granulomatous retinitis, choroiditis, and endophtalmitis.[3,4] Although rare, there are reports of blindness by choroiditis and enucleation due to endophthalmitis. In general, in cases of intraocular involvement, there is associated immunosuppression, often with exuberant skin lesions and involvement of other sites by dissemination.[5,6] Thus, it should be noted that all patients with disseminated sporotrichosis and immunosuppression should be referred to an ophthalmologist for early detection of possible eye involvement.

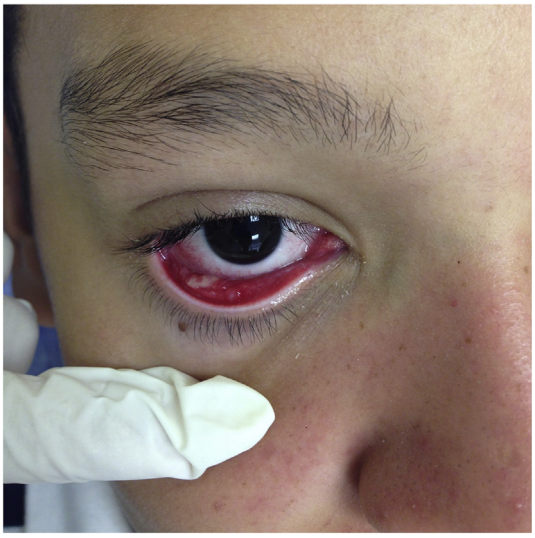

Fig. 6. Patient 3 - Inferior tarsal granulomatous conjunctivitis of the right eye.

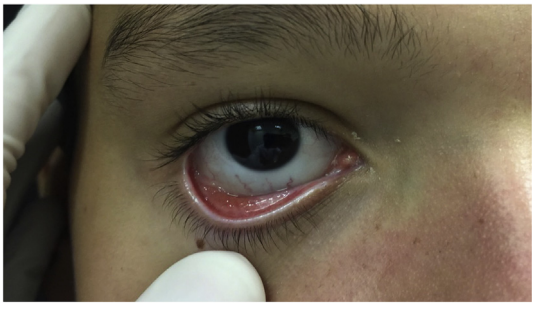

Fig. 7. Patient 3 - Resolution of conjunctiva lesions 15 days after initiation of treatment.

On the other hand, ocular sporotrichosis involving only the ocular adnexa can manifest as granulomatous conjunctivitis, Parinaud's oculoglandular syndrome (POS), dacryocystitis, and bulbar conjunctivitis. In granulomatous conjunctivitis, grouped yellowish nodules are observed, with a smooth, shiny surface involving the tarsal and/or bulbar conjunctiva, associated with conjunctival hyperemia and/or purulent exudate.[7] Initially, the nodule caused by granulomatous conjunctivitis is often confused with hordeolum and chalazion, and ocular sporotrichosis is an important differential diagnosis in lesions not responding to standard treatment. POS occurs when the patient has granulomatous conjunctivitis and preauricular or submandibular ipsilateral lymphadenopathy, as observed in patients 2 and 3.[8] Acute dacryocystitis is an inflammation of the lacrimal sac with infiltration, swelling, and induration observed in the medial canthal ligament of the eye and can progress to become chronic.[9] Bulbar conjunctivitis is when this mucosa is infiltrated, with pink coloration and hiperemia.[10]

Although infrequent, most cases of ocular sporotrichosis affect the ocular adnexa. According to work published by Schubach et al. (2005)[7] of sporotrichosis cases, only 2.3% had conjunctival lesions, with 0.7% showing primary conjunctival involvement, without skin lesions. Evolution is generally benign and rarely produces sequelae, such as fibrosis and chronic dacryocystitis. A recent literature review recorded 65 cases of ocular sporotrichosis, mostly in highly endemic areas (Peru and Brazil), with 21 new cases in the Abancay region of Peru, with a high frequency of palpebral involvement (82%).[11]

The transmission of sporotrichosis was always thought to occur through traumatic implantation of *Sporothrix* sp. conidia from a contaminated source, such as decomposing vegetables and soil. The state of Rio de Janeiro, Brazil, faces a sporotrichosis epidemic since 1998 of zoonotic transmission involving domestic and stray cats. This transmission usually occurs by cat scratches or bites.[12] However, in addition to traumatic implantation, there are rare cases of pulmonary and disseminated sporotrichosis acquired through inhalation.[13] Other studies have reported cases of sporotrichosis developed after manipulation of a contaminated source, with no history of traumatic inoculation.[14] It is believed that the high virulence of *Sporothrix brasiliensis* (the most prevalent species in the endemic area of Rio de Janeiro) is the reason for the increase in atypical cases and atraumatic inoculation of the fungus.[2,15]

In the three cases reported here, there was no ocular trauma. However, patient 1 was a gardener, and patients 2 and 3 lived with ailing cats. Contact with ailing animals appears to be sufficient for human exposure to the fungus and for self-inoculation. Sick cats have lesions richly parasitized by *Sporothrix brasiliensis* and many develop respiratory symptoms, such as sneezing, increasing the risk of transmission of the fungus.[12]

The approach to a case of ocular sporotrichosis is very similar to that for the cutaneous form. Diagnosis is made by collection of the conjunctival discharge with a sterile swab and culturing the material for fungi. Because of its association with disseminated sporotrichosis, the intraocular form should be treated with amphotericin B as a loading therapy, and itraconazole as maintenance, except for endophthalmitis, which should be treated with intravitreal and/or intravenous amphotericin B.[16] Sporotrichosis affecting the ocular adnexa should be treated with itraconazole in the same dose as the cutaneous form: 100–200 mg/day until complete resolution of the lesions[12] (or for 2-4 additional weeks),[16] in general a total of 3–6 months.[12,16]

Although in case 3, treatment evolved as expected, with efficacy and without sequelae, the other two patients required time and/or dose adjustments to their regimens. This delay in the response to treatment may have been due to the late start of the treatment of the first patient and the extensive ocular and periocular involvement of the second patient.

4. Conclusions

Very few case reports of ocular sporotrichosis are available. Although the incidence of this type of sporotrichosis is increasing in epidemic areas, clinical similarities with other types of conjunctivitis may mislead the ophthalmologist to treat cases of ocular sporotrichosis with antibiotics and postponing definite cure. We emphasize the importance of the ophthalmologist being familiar with the diagnosis and management of this rare and frequently misdiagnosed form of sporotrichosis.

Patients consent

Written consent for publication of personal identifying information including medical record details and photographs was obtained from all three patients.

IRB Approval: The Research Ethics Committee of the Evandro Chagas National Institute of Infectious Diseases, Fiocruz, RJ, Brazil, approved this study under the protocol number 08097112.3.0000.5262. All patients involved were anonymized for privacy and ethics purposes.

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Conflict of interest

All the authors have no financial disclosures.

Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

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