



Ocular and cutaneous sporotrichosis

Aluisio Rosa Gameiro Filho^{a,*}, Carolina Tagliari Estacia^a, Rodrigo Rosa Gameiro^b,
Leandro de Mattos Fonseca Vieira^a, Daniella Socci da Costa^a

^a Hospital Federal dos Servidores do Estado do Rio de Janeiro, Rua Sacadura Cabral, 178, Neighborhood: Saúde, Rio de Janeiro, Rio de Janeiro, 20221-903, Brazil

^b Pontifícia Universidade Católica do Paraná, Rua Imaculada Conceição, 1155, Neighborhood: Prado Velho, Curitiba, Paraná, 80215-901, Brazil

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ABSTRACT

Purpose: Sporotrichosis is the most common subcutaneous mycosis in the world and it is presented usually in the lymphocutaneous form. When the eye is involved, it typically includes the conjunctival and periorbital tissues or even the intraocular tissues.

Observations: We present a case of a 13-year-old girl complaining about nodules in the left malar region and in both the outer nasal corner and the lower eyelid of the left eye, which had appeared one month earlier.

Conclusions: and significance: Sporotrichosis is a hyperendemic disease in Brazil and its diagnosis is based on clinical aspects, epidemiological history, and culture. Ophthalmologists and other professionals should be aware of this condition.

1. Introduction

Sporotrichosis is the most common subcutaneous mycosis in the world, especially in regions of tropical or subtropical climate.² It is characterized by nodular cutaneous or subcutaneous lesions, with adjacent prominent lymphatic vessels. It is caused by the dimorphic fungus *Sporothrix schenckii*, usually found in soil and other vegetable matters, and penetrate the organism by either traumatic implantation into the skin or, rarely, by inhalation. In Brazil, the disease is transmitted mostly through bites or scratches of felines contaminated with the fungus. Handling the lesion (e.g., scratching) may lead to secondary contamination and delayed diagnosis. *S. brasiliensis* is the main etiological agent of the highest feline zoonotic transmission reported in southeastern Brazil since the late 1990s¹.

Ocular involvement is rare and most reports describe the involvement of the conjunctival and periorbital tissues (presented in only 2.3% of all sporotrichosis cases),¹ which is usually associated with direct facial trauma. Intraocular presentation is rare and a common consequence of hematogenous dissemination from multiple systemic lesions.²

We report a case of a 13-year-old girl who sought assistance at the Ophthalmology Department of the Hospital Federal dos Servidores do Estado do Rio de Janeiro, complaining about facial skin nodules that had appeared one month earlier.

1.1. Case report

A 13 years old black healthy female patient, residing in the metropolitan area of Rio de Janeiro, RJ, Brazil complained about nodules in the left malar region that had appeared one month earlier, followed by a similar lesion in nasal area of the left upper eyelid and also in the lower eyelid of the left eye. She referred a progressive growth, as well as previous ophthalmological evaluations at other hospitals for the same issues. The patient also reported to have already used oral cephalixin for 10 days, as well as topical dexamethasone associated with neomycin and polymyxin B sulfate, and gentamicin ointment, but without improvement.

The slit-lamp examination showed, in the left eye, follicles in the lower tarsal conjunctiva, conjunctival hyperemia, lower eyelid swelling, and a granulomatous grayish lesion that drained a brownish exudate. The right eye and the fundus examination of both eyes were normal. The best-corrected visual acuity (BCVA) was 20/20 in both eyes.

The clinical examination showed the presence of a nodular lesion in the inner corner of the left eye measuring 1 × 1 cm (cm), and a lesion in the left lower eyelid measuring 3.5 × 2 cm, with a granulomatous appearance and dissemination to the lymphatic chain. There was also a similar lesion in left malar region, measuring 2.5 × 2 cm. The patient also presented painless ipsilateral lymphadenopathy in both preauricular and anterior cervical chains (Fig. 1A–C Fig. 2 and 2A–D). Lacrimal apparatus was not involved.

* Corresponding author. Rua Vinícius de Moraes, 100 apartment 608, Ipanema, Rio de Janeiro – RJ, Brazil.

E-mail address: gameirofilhomd@gmail.com (A.R. Gameiro Filho).

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Fig. 1. Fig. 1A: The patient presented a nodular lesion in the inner corner of the left eye, measuring 1 × 1 cm; a lower eyelid granulomatous lesion, measuring 3.5 × 2 cm, and a similar lesion in the left malar region, measuring 2.5 × 2 cm/1B: Cervical lymphadenopathy/1C: Facial and ocular granulomatous lesions/The cutaneous and eyelid lesions show complete resolution 3 months after Itraconazole treatment (D, E). Patient remained with a residual scar in her malar region (F).



Fig. 2. Fig. 2A: Granulomatous lesion in the lower eyelid of the left eye/B: Involvement of inferior tarsal conjunctiva of the left eye, showing follicles/C: Inner corner nodular lesion/D: Granulomatous malar lesion/The cutaneous and eyelid lesions show complete resolution 3 months after Itraconazole treatment (E, F, G). Patient remained with a residual scar in her malar region (H).

The patient presented low socioeconomic status and reported upon inquiry to have adopted a cat from the streets before the symptoms started. She mentioned the cat was ill and had died a few weeks before the medical appointment reported in this study. She did not remember of any injuries or scratches of the cat in her face.

Considering the epidemiological history and the clinical appearance of the lesions, the diagnosis of sporotrichosis was suspected. A skin

biopsy was performed and the specimens were sent for histological analysis. The culture was positive for *Sporothrix brasiliensis*, appearing on SGA (Sabouraud glucose agar) at 25 °C as a delicate branching, molded with pyriform conidia, in a typical flower-like arrangement. The diagnosis of ocular and cutaneous sporotrichosis was established and 200 mg/day of Itraconazole was prescribed for 3 months. The patient presented a gradual and satisfactory improvement until complete

resolution, and only a small scar remained in the left malar region and in the lower eyelid margin of the left eye after the treatment (Fig. 1D–F and 2E–H).

2. Discussion

Ocular sporotrichosis is usually presented as a granulomatous conjunctivitis,¹ in which nodular yellowish elevations involving bulbar or tarsal conjunctiva are present and are often rather difficult to distinguish from follicles.² These nodules are grouped and feature a smooth surface, and may be confused with hordeolum and chalazion, delaying the diagnosis in some cases.³ They may be associated with conjunctival hyperemia and mucopurulent exudate.⁴ Parinaud's oculo-glandular syndrome occurs when granulomatous conjunctivitis is associated with ipsilateral lymphadenopathy, usually involving preauricular, submandibular, and anterior cervical chains.⁵

Other possible presentations are bulbar conjunctivitis with mucosal infiltration,⁶ which may be confused with adenoviral conjunctivitis⁷ and even MALT lymphoma⁸; episcleritis; acute or chronic and persistent dacryocystitis⁹; fungal keratitis, corneal ulceration; and ectropion.

The intraocular involvement may be presented as granulomatous uveitis; iridocyclitis; choroiditis, which can be multifocal⁹; retinal granulomas¹⁰; and endophthalmitis, which may lead to amaurosis.¹¹ This type of involvement is usually associated with immunosuppressive conditions such as HIV.

We believe that our patient had self-inoculated the fungus when scratching or handling the lesion, which destabilized it^{12 13} and may explain the appearance of multiple lesions. Our patient also used topical steroids as previous treatment, which might have facilitated fungus replication, modifying the morphology of the lesion and making it more exuberant.

For diagnosis, clinical suspicion is the key and culture is the gold standard. Among other tests available, histopathology is nonspecific and may mimic other granulomatous conditions. Intradermal or molecular testing, such as PCR, may also be performed, but they usually present variations of sensitivity and specificity⁶ and are not available in most hospitals.

The treatment is similar to the cutaneous form of the disease and it should start as soon as possible to prevent sequelae such as symblepharon and scars. The gold standard¹⁴ is the use of Itraconazole, which should be administered orally in a dose of 200–400 mg/day for 3–6 months (or for 2–4 weeks in addition to the resolution of lesions). Patients using steroids or with immunosuppressive status might benefit from a prolonged treatment time. The most important disadvantage of this drug is the high cost, especially in developing countries, where sporotrichosis is more prevalent.¹⁵ Fluconazole is another potential azole to be used.

Our patient had a complete improvement of her lesions with 3 months of Itraconazole, remaining only a small scar in the left malar region and in the lower eyelid margin of her left eye.

3. Conclusions

Ocular sporotrichosis may be similar to other eye conditions, which can be confusing and challenging to the ophthalmologist, usually delaying diagnosis. It may also postpone treatment and lead to sequelae, as it occurred in the present case. Hence, both the physician and the ophthalmologist should be aware of this condition in order to start the correct treatment as soon as possible.

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Intellectual property

We confirm that we have given due consideration to the protection of intellectual property associated with this work and that there are no impediments to publication, including the timing of publication, with respect to intellectual property. In so doing we confirm that we have followed the regulations of our institutions concerning intellectual property.

Patient consent

Written consent to publish potentially identifying information, such as details or the case and photographs, was obtained from patient's legal guardian.

Authorship

All listed authors meet the ICMJE criteria. We attest that all authors contributed significantly to the creation of this manuscript, each having fulfilled criteria as established by the ICMJE.

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Declaration of competing interest

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