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Case report

Ocular Sporotrichosis

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ABSTRACT

Purpose: To present 10 cases of Parinaud oculoglandular syndrome caused by sporotrichosis.
Observations: We report 10 cases of Parinaud oculoglandular syndrome after contact with domestic cats diagnosed with sporotrichosis. They all showed ocular hyperemia associated with unilateral tarsal conjunctival granulomas. After histopathological study and culture of the scrapings and conjunctival secretions, six patients were positive for *Sporothrix schenkii* and four had a presumed diagnosis of Parinaud oculoglandular syndrome due to sporotrichosis. Treatment with 200 mg/day of oral itraconazole was started, and all patients had a favorable therapeutic response.
Conclusion and importance: These case reports are essential for characterizing a rare etiology of Parinaud oculoglandular syndrome.

1. Introduction

Sporotrichosis is caused by *Sporothrix schenckii,* a dimorphic fungus responsible for an infection that is typically localized to the skin and subcutaneous tissue.[1] Infection usually occurs due to traumatic inoculation of vegetables, soil, or organic material contaminated with the fungus. Zoonotic transmission is also possible, but has been described only in isolated cases or small outbreaks.[2]

The cutaneous form of sporotrichosis represents the most common clinical presentation of the disease[3]; while the extracutaneous form is less prevalent and ocular involvement has rarely been described in immunocompetent patients, or in individuals without previous ocular trauma.[4]

Sporotrichosis may manifest as Parinaud oculoglandular syndrome, which is characterized by granulomatous conjunctivitis associated with preauricular and submandibular lymphadenopathy.[5]

This paper presents 10 cases of Parinaud oculoglandular syndrome caused by sporotrichosis, diagnosed considering clinical features, epidemiological findings, culture and histopathological data. We describe six positive culture cases (1–6) and four cases with presumed diagnosis (7–10).

2. Findings

2.1. Case 1

A 69-year-old housewife complained of conjunctival hyperemia, itching, edema in the right eye (OD), and nodules in the right malar region. She denied a history of trauma but reported that, a few weeks prior, her cat started to exhibit bodily injuries and died due to sporotrichosis. The best-corrected visual acuity was 20/60 in the OD and 20/30 in the left eye (OS). Examination revealed erythematous nodules with slight perilesional infiltration in the right malar region ascending to the ipsilateral upper lid. Some nodules were ulcerated and with abscess formation. Biomicroscopy revealed conjunctival hyperemia, follicles, and several granulomas in the upper and lower eyelid. Fundoscopy was normal.

2.2. Case 2

A 13-year-old female student was referred by her mother. She presented with ocular hyperemia, periocular edema, and some nodules in the OD inferior tarsal conjunctiva that had been apparent for several weeks. She denied a history of trauma and reported that their cat had

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been diagnosed with sporotrichosis. Examination showed a best-corrected visual acuity of 20/20 and right preauricular and submandibular lymphadenopathy. Biomicroscopy showed mild upper eyelid edema in the OD, conjunctival hyperemia 1+/4+, several granulomas in the right lower eyelid, and in the right upper eyelid (Fig. 1-A). Serous secretion was present in the fornix, and the patient had a transparent cornea with no anterior chamber inflammatory reaction, as per the first case.

2.3. Case 3

A 22-year-old male biologist complained of pain and ocular hyperemia in the OD for 1 week. He denied a history of trauma. Examination showed a best-corrected visual acuity of 20/20 and right preauricular and submandibular lymphadenopathy. The patient showed progressive worsening of his ophthalmic condition, characterized by upper eyelid edema, conjunctival hyperemia (3+/4), chemosis (3+/4), a granulomatous lesion near the caruncle, mucopurulent secretion, and papillae in the tarsal conjunctiva (Fig. 1-B). He was treated for gonococcal conjunctivitis with intramuscular ceftriaxone and topical moxifloxacin but showed no improvement. The possibility of ocular spotrichosis was then suggested after the patient reported that his cat have been diagnosed with sporotrichosis.

2.4. Case 4

A 18-year-old female student complained of ocular hyperemia associated with headache and a febrile sensation for 2 weeks. She denied any history of trauma. She reported that a few weeks prior, her cat exhibited bodily ulcerations, and had been diagnosed with sporotrichosis. Examination showed uncorrected visual acuity of 20/20 and right submandibular lymphadenopathy (Fig. 1-C). Biomicroscopy showed mild upper eyelid edema in the OD, conjunctival hyperemia 1+/4+, several granulomas in the right lower eyelid, one granuloma in the nasal region of the right upper eyelid, and several granulomas in the temporal region of the right upper eyelid. Serous secretion was present in the fornix, and the patient had a transparent cornea with no anterior chamber inflammatory reaction.

2.5. Case 5

A 21-year-old female veterinary student complained of conjunctival hyperemia and some nodules in the OS. She denied a history of any other trauma but reported that her cat had been diagnosed with sporotrichosis. The best -corrected visual acuity was 20/20 in the OD and 20/30 in the OS. Biomicroscopy revealed conjunctival hyperemia (1+/4+), granulomas in the inferior tarsal conjunctiva with serous secretion in the fornix, a transparent cornea, and no anterior chamber inflammatory reaction in the OS (Fig. 1-D).

2.6. Case 6

A 15-year-old male student complained of conjunctival hyperemia in the OS for 8 days. He denied a history of trauma but reported that his cat

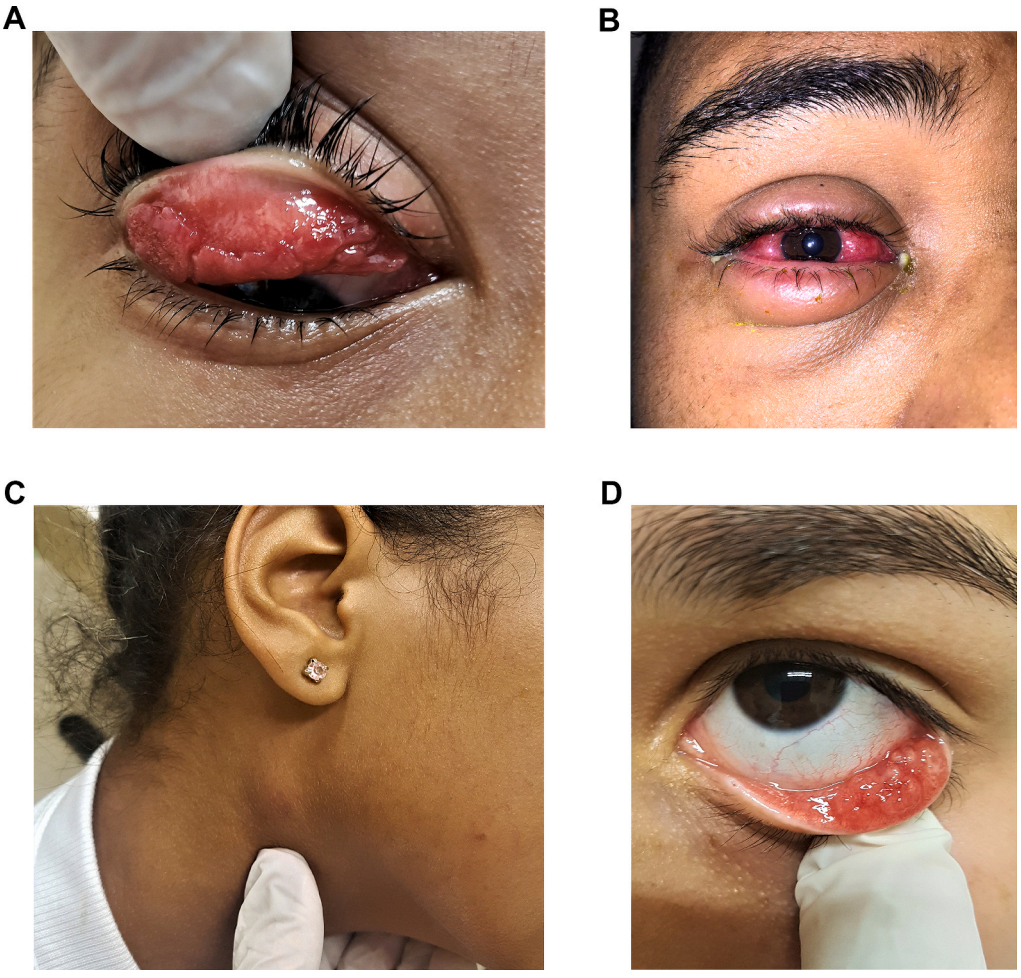

Fig. 1. Patients with positive culture for *Sporothrix schenckii*. Granulomas in the upper right palpebral conjunctiva (A). Periorbital edema on the right side with infiltration of the bulbar conjunctiva (B). Submandibular lymphadenopathy (C). Granulomas in the lower left palpebral conjunctiva (D).

had died from sporotrichosis. Physical examination revealed left preauricular and submandibular lymphadenopathy and visual acuity of 20/20. Biomicroscopy showed conjunctival hyperemia 1+/4+, a granulomatous lesion in the inferior tarsal conjunctiva, and papillae in the tarsal conjunctiva with serous secretion in the fornix, transparent cornea, with no anterior chamber reaction in the OS.

After histopathological study and culture of the scrapings and conjunctival secretions, all the six patients above were positive for *Sporothrix schenkii* (Fig. 2). Treatment with 200 mg/day of oral itraconazole for 3–6 months was started, and all patients had a favorable therapeutic response after the third month.

2.7. Cases 7–10

Four patients (three female and one male) complained of chronic conjunctival hyperemia. They reported that their cats have been diagnosed with sporotrichosis. They all denied a history of trauma. Dermatologic examination showed ipsilateral preauricular and submandibular lymphadenopathy. Ophthalmology examination showed corrected visual acuity of 20/20. Biomicroscopy showed conjunctival hyperemia, granulomas in the eyelid with serous secretion in the fornix, a transparent cornea, and no anterior chamber inflammatory reaction.

Histopathological studies and cultures of scrapings and conjunctival secretions were performed and, although patients 7 to 10 were negative for *Sporothrix schenkii*, their clinical presentation and epidemiological findings suggested Parinaud oculoglandular syndrome due to sporotrichosis. They were treated with 200 mg/day itraconazole because of their presumed diagnoses and they all showed a favorable therapeutic response.

3. Discussion

Since 1980, zoonotic transmission of sporotrichosis has been mentioned in reports, usually in the setting of outbreaks in the home involving animals, their owners, and/or veterinarians.[2] In the home environment, sporotrichosis usually occurs transmitted through biting, scratching, or contact with the secretions of infected animals, especially cats.[1] In the 10 cases reported, the patients denied eye trauma; however, they all had a pet cat diagnosed with sporotrichosis, which was probably the source of contamination.

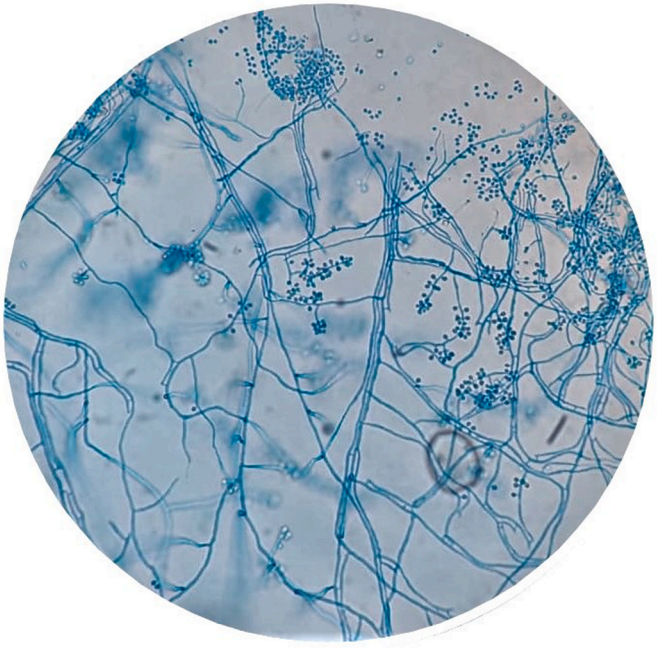

Fig. 2. Photomicrograph of *Sporothrix schenckii*.

Most of the patients in the present study were female. There are epidemiological data pertaining to occupation, sex, and routes of transmission showing that sporotrichosis is predominant in adult women engaged in domestic activities, probably because this group is more exposed to the fungus due to close contact with pets under their care.[6]

Brazil is an endemic area for sporotrichosis.[6] The clinical presentations of sporotrichosis are diverse and medical studies differ regarding their classification of the disease.[7] The most accepted classification distinguishes the disease into cutaneous and extra-cutaneous clinical forms, with the former being the most common form, accounting for 80% of all cases.[8] Although it is rare, most cases of ocular sporotrichosis show involvement of ocular adnexa, with primary conjunctival involvement without skin lesions affecting only 0.7% of all sporotrichosis cases, according to Schubach et al. (2005).[3] In the present study, this was observed in 90% of the cases.

Very few case reports of ocular sporotrichosis are available. However, the incidence is increasing in endemic areas. Clinical similarities with other forms of conjunctivitis may lead ophthalmologists to initiate antibiotic treatment, thus postponing definitive treatment.[9]

Sporotrichosis can be diagnosed on the basis of a combination of clinical, epidemiological, and laboratory data.[2]

Parinaud oculoglandular syndrome is characterized by the clinical presentation of unilateral granular or tarsal granulomatous conjunctivitis with or without painful ipsilateral regional lymphadenopathy; it may also present with adjacent systemic symptoms such as fever, conjunctival follicular reaction, foreign body sensations, and hyperemia. Although classically associated with *Bartonella hanselae* infection, other etiologies have been identified, such as viruses, fungi, parasites and mycobacteria, although these are less common.[10]

Laboratory tests for the determination of sporotrichosis include direct analysis of specimens, such as tissue biopsy specimens or lesion secretions. In cases with disseminated infection, other specimens may also be analyzed, such as sputum, urine, blood, or synovial fluid.[2]

Culture is the gold standard for establishing the diagnosis of sporotrichosis.[11] Generally, Sabouraud dextrose agar is used for primary isolation.[1] In the present study, this was the medium we used.

Sporothrix schenckii was found to be a complex of species, including *Sporothrix brasiliensis*[12]; which has been implicated in the hyperendemic transmission of sporotrichosis in Brazil. A molecular analysis of the strains in these ten cases was not performed; however, based on epidemiology, it is likely that *S. brasiliensis* was the species involved.

Intradermal and molecular tests are of low diagnostic value for sporotrichosis because of their high variability in sensitivity and specificity; they are also less likely to be routinely available. Nevertheless, these tests can be useful in terms of raising diagnostic suspicion and thus prompting a more aggressive diagnostic approach.[11]

The ELISA shows a sensitivity of 90% and specificity of 86%. This diagnostic tool can be used in both typical and atypical cases of sporotrichosis, and for patients in whom mycological studies were negative. Serological testing is useful for therapeutic follow-up.[13]

Spontaneous resolution of sporotrichosis is extremely rare and most patients require drug intervention. Regarding treatment, several drugs have been used previously, including potassium iodide, which was the first treatment to be applied and the drug of choice for a long time. However, potassium iodide is not effective in extracutaneous forms of the disease.[14] Currently, itraconazole, an oral antifungal agent of the azoles class, is the drug of choice for treating sporotrichosis.[2,11,14] For cutaneous and lymphocutaneous sporotrichosis, itraconazole (200 mg taken orally each day) is recommended. The treatment duration may vary from 2 to 4 weeks after the disappearance of all lesions and it usually takes 3–6 months.[14] In our 10 cases, a dose of itraconazole of 200 mg/day was used for 3–6 months, and a favorable therapeutic response was achieved after the third month in all cases.

4. Conclusions

Case reports are essential for characterizing rare etiologies of Parinaud oculoglandular syndrome, which is becoming increasingly common, especially in developing countries such as Brazil. Diagnostic suspicion is essential to reduce the risk of morbidity and to relieve the symptoms as soon as possible. Clinical history taking is critical in all cases, and contact with pets, healthy or otherwise, should not be overlooked. Further studies on zoonotic transmission of sporotrichosis and the possible ocular and systemic manifestations of this etiology are urgently needed, given the increasing number of cases in recent years.

Patient consent

Written informed consent was obtained from patients for publication of these case reports and any accompanying images.

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Authorship

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

Declaration of competing interest

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