

Primary ocular sporotrichosis with granulomatous conjunctivitis

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Abstract

Sporotrichosis is an infection which is caused by the dimorphic fungus *Sporothrix schenckii*. Primary ocular sporotrichosis is uncommon in non-endemic areas and may be easily misdiagnosed, leading to a delay in initiation of treatment. In this case report, a 15-year-old girl, who is a post renal transplant patient presented with left eye swelling and localized redness at the medial canthal region. She was initially treated with topical antibiotics but there was no improvement. The addition of topical steroids led to the development of multiple nodules with central ulceration. Examination of the left eye showed granulomatous conjunctivitis. The features were suggestive of sporotrichosis and she was empirically started on oral itraconazole. A biopsy of the lesion showed caseating granulomatous inflammation and fungal PCR tested positive for *Sporothrix schenckii*. Her symptoms and clinical findings completely resolved after 3 weeks on itraconazole; however, her liver function deteriorated, and patient opted to discontinue the medication.

areas.¹ *Sporothrix* constitutes a species complex that includes *Sporothrix schenckii*, *Sporothrix brasiliensis*, *Sporothrix globosa*, *Sporothrix mexicana*, *Sporothrix albicans*, and *Sporothrix lurei*.² It is commonly found in soil and decaying vegetation. Infection in humans usually occurs by traumatic inoculation of the fungus.

Classification of sporotrichosis is broadly divided into two categories (cutaneous and extracutaneous sporotrichosis), which comprise four distinct clinical forms: lymphocutaneous, fixed cutaneous, disseminated cutaneous, and extra-cutaneous.³ The most frequent clinical presentation of sporotrichosis is the lymphocutaneous form (55%).⁴ Ocular sporotrichosis is uncommon in non-endemic areas and has rarely been described in individuals without prior ocular trauma. We report a rare case of primary ocular sporotrichosis affecting the bulbar and palpebral conjunctiva with no preceding trauma.

Introduction

Sporotrichosis, an infection which is caused by dimorphic fungus *Sporothrix schenckii*, is prevalent worldwide, but particularly in tropical and subtropical

Case Report

A 15-year-old girl who is post renal transplant on long term immunosuppression therapy presented with left eye swelling at the medial canthal region and localized redness for the past 1 month (Figure 1). It was associated with minimal eye discharge, with no blurring of vision. There was no preceding fever, skin

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lesions or respiratory symptoms. She has 3 pet cats at home which were all vaccinated. There was no



Figure 1
Initial clinical presentation prior to diagnosis of ocular sporotrichosis and prior to treatment demonstrated mild periorbital edema and generalized conjunctival injection with swelling at the medial canthal region of the left eye.

history of cat scratch, trauma or contact with organic matter.

She was initially treated with topical antibiotics, however there was no improvement and the lesion progressively increased in size. The addition of topical steroids led to development of multiple nodules with central ulceration over the bulbar and forniceal conjunctiva. During this period, she also developed a string of palpable masses adjacent to her left nasolabial fold (Figure 2).

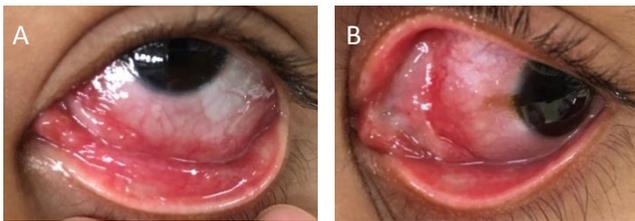


Figure 2
Granulomatous conjunctival lesions with overlying ulceration at the medial canthal region (A) and lower tarsal conjunctiva (B), in the left eye.

Upon presentation for ophthalmic examination, her vision was 20/20 in each eye. Slit-lamp biomicroscopy of the left eye showed generalized conjunctival injection and a fleshy mass with overlying ulceration at the medial canthal region. Similar ulcerated granulomatous lesions were present on upper and lower tarsal conjunctiva. Right eye anterior segment and bilateral fundus examination were unremarkable. A cord like mass on the left

cheek was palpable, which was non tender with no overlying skin changes. There was no preauricular lymphadenopathy.

She was initially treated as presumed ocular sporotrichosis and empirically started on 200 mg itraconazole orally once per day. Fungal culture was negative and *Sporothrix schenckii* was not detected in histopathological examination. An incisional biopsy of the conjunctival lesion later revealed caseating granulomatous inflammation (Figure 3) and fungal PCR tested positive for *Sporothrix schenckii*. Her

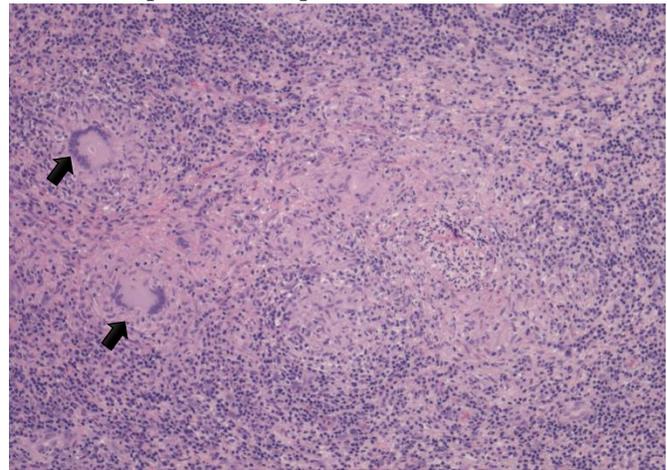


Figure 3
Histopathology of the conjunctival lesion stained with hematoxylin and eosin disclosed collections of non-caseating granulomas with Langhans multinucleated giant cells denoted by the arrows (at 100x).

condition improved gradually and the conjunctival lesions including the cord like mass at the left cheek completely resolved (Figure 4). Her symptoms and clinical findings completely resolved after 3 weeks on itraconazole; however, her liver function deteriorated, and patient opted to discontinue the medication.



Figure 3
Clinical presentation following 3 weeks of treatment.

Discussion

Sporotrichosis is a subacute or chronic mycosis, mainly involving the cutaneous tegument of infected patients. Primary ocular sporotrichosis is rare and can occur in two forms which are intraocular or as an external eye disease.⁵ The intraocular form occurs via hematogenous dissemination. It is usually associated with immunosuppression, presenting as anterior or posterior uveitis, choroiditis, retinal granuloma, or endophthalmitis. On the other hand, external form occurs via auto-inoculation which may present as conjunctivitis, scleritis, episcleritis, corneal ulcer, dacryocystitis, or lid granulomas.^{4,6-8} There can also be simultaneous involvement of the conjunctiva and the regional lymph nodes, which was seen in our patient and is known as Parinaud syndrome.⁹

Classical sporotrichosis infection is associated with traumatic subcutaneous inoculation of soil, plants, or organic matter contaminated with fungus. Though zoonotic transmission is rare, there has been an increasing number of cases reported whereby patients were infected after contact with cats.¹⁰⁻¹³ Sick cats typically harbour a high yeast-like fungal burden and can transmit the disease via cat scratches and bites, or by non-traumatic ways, such as a cat's cough or sneezing with direct contact between patients' integumental barriers and animal secretions.¹⁴

In Malaysia, *Sporothrix schenckii* is the prevailing causative agent of feline sporotrichosis.¹⁵ In this case, the patient's conjunctival biopsy for fungal PCR also tested positive for *Sporothrix schenckii*. This is in contrast with cases reported in Brazil where the primary pathogen of feline sporotrichosis is *Sporothrix brasiliensis*.^{9,16,17}

Recognizing and diagnosing ocular sporotrichosis is often challenging due to the variable ocular presentation. Several case reports have shown that ocular sporotrichosis is frequently misdiagnosed due to its rarity in non-endemic regions and diverse clinical presentation, which mimics other eye conditions.^{6,12,18} A high index of suspicion is important to ensure proper treatment is administered in a timely manner. This high index of suspicion is especially vital in

immunocompromised patients such as our patient, as sporotrichosis in immunocompromised hosts may harbour a more severe clinical course. Queiroz-Telles, *et al.* reported different forms of sporotrichosis in post renal transplant patients which included cutaneous, osteoarticular, pulmonary and disseminated form but none with solely ocular involvement.³

The gold standard for establishing the diagnosis of sporotrichosis is through a positive culture. Histopathologic examination of tissue samples will reveal a mixed granulomatous and pyogenic inflammatory process, but the organisms are often difficult to visualize due to the small number of organisms that are often present. In our patient, *Sporothrix schenckii* was not detected on fungal culture nor on histopathologic examination. However, fungal PCR of the conjunctival biopsy tested positive for *Sporothrix schenckii*.

The main aim of treatment is to achieve resolution of active infection and eradication of *Sporothrix schenckii* from tissues. Several treatment options are available which include azoles such as itraconazole, saturated solution of potassium iodide (SSKI), amphotericin B, and allylamines such as terbinafine.^{9,19,20} The treatment duration of sporotrichosis depends on the clinical form. In a study conducted by Arinelli, *et al.* the suggested treatment regime for ocular sporotrichosis is oral itraconazole, with an initial dosage of 100 mg per day for a minimum period of 90 days, and discontinuation after having a full month of ocular disease resolution.⁸ Therapy of immunosuppressed patients with sporotrichosis does not differ from treatment modalities in immunocompetent individuals.³ Our patient achieved complete clinical resolution within 3 weeks of treatment with itraconazole, but she opted to discontinue the medication due to deteriorating liver function.

Conclusion

Ocular sporotrichosis with primary conjunctival involvement without skin lesions is rare. The similarity with other clinical conjunctivitis may lead

to a delay in treatment and, as a consequence, increase risk of sequelae in the eye and risk of disseminated disease particularly in hosts with impaired host defences. Thus, a high index of suspicion and early treatment is crucial in combating this entity.

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