A CASE REPORT AND LITERATURE REVIEW ON GRANULOMATOUS CONJUNCTIVITIS AND LYMPHOCUTANEOUS LESIONS ASSOCIATED WITH SPOROTRICHOSIS

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Abstract

Sporotrichosis, a skin mycosis caused by *Sporothrix sp*., is a rarely reported condition in Malaysia. In this case report, we present a case of granulomatous conjunctivitis and lymphocutaneous lesions caused by sporotrichosis, along with a review of the relevant literature available on PUBMED. A 70-year-old woman presented with mucopurulent discharge from her left eye and nodular skin lesions on her face. She reported previous contact with her cat, which had recently died from sporotrichosis. Upon examination, the left eyelid was edematous, and there was conjunctival hyperemia. Multiple granulomatous lesions were observed in the bulbar conjunctiva, covered by the mucopurulent discharge. Additionally, erythematous and granulomatous nodules were present on the left periorbital area, extending to the left malar area, accompanied by ipsilateral lymphadenopathy. Based on clinical suspicion, ocular and lymphocutaneous sporotrichosis was diagnosed. Treatment was initiated with oral itraconazole and topical fluconazole empirically. The conjunctival sample tested negative for *Sporothrix schenckii*. Histopathological examination of skin biopsy specimens revealed nonspecific granulomatous inflammation. After three months of antifungal treatment, the patient recovered completely without any ocular complications.

Keywords: Ocular Sporotrichosis, Granulomatous Conjunctivitis, *Sporothrix schenckii*

Introduction

Sporotrichosis is a cutaneous mycosis commonly reported in endemic areas like Peru and Brazil. It is caused by the dimorphic fungus *Sporothrix spp.* (1). The lymphocutaneous form is the commonest form of presentation, representing 80% of cases reported. Ocular involvement is typically less prevalent. Conjunctival lesions are found in 2.3% of cases, with 0.7% showing primary ocular involvement without a skin lesion (2). Here we describe a case of granulomatous conjunctivitis and lymphocutaneous lesions due to sporotrichosis which exhibited a good response to antifungal treatment.

Method

The methodology employed for this study involved a case report along with a review of the relevant literature available on PUBMED.

Case description

A healthy 70-year-old housewife suffered a month of left-eye mucopurulent discharge associated with nodular skin lesions on the left periorbital area, cheek, and neck. She was initially treated in a private hospital for left-sided bacterial conjunctivitis. Topical antibiotics and steroids were administered but did not show any improvement. In addition, a skin biopsy was also done to rule out malignancy. There was no history of prior trauma or contact with organic matter. However, the patient disclosed that she had a cat residing at her home before this incident, although she denied being scratched or bitten by the cat. The patient later disclosed that her cat had succumbed to sporotrichosis approximately a month prior when she took it to a veterinarian after noticing multiple ulcerating lesions with discharge on the pinnae, nose, and head.

Ophthalmic examination revealed visual acuity of 6/15 in both eyes. The left eyelid exhibited swelling along with

conjunctival hyperemia. Multiple granulomatous lesions were observed in the bulbar conjunctiva, covered by mucopurulent discharge (Figures 1a and 1b). Otherwise, the left eye cornea is clear, and no anterior chamber inflammatory reaction exists. The posterior segment finding was unremarkable. Dermatological examination showed erythematous, granulomatous nodules over the left periorbital area extending to the left malar region, accompanied by lymphadenopathy in the ipsilateral preauricular, submandibular, and cervical regions. No skin ulcers or abscesses were present (Figure 2). Based on clinical suspicion, she was diagnosed with ocular and lymphocutaneous sporotrichosis. Treatment was initiated with oral itraconazole (400 mg/day) and topical fluconazole (0.2% QID). A conjunctival swab was sent for culture and sensitivity, and the patient was referred to a dermatologist for the evaluation of skin lesions. The skin biopsy showed non-specific granulomatous inflammation, while the conjunctival swab later tested negative for *Sporothrix schenckii.* Despite this, the patient exhibited clinical improvement with reduced eye redness and resolution of skin lesions after two weeks of systemic antifungal treatment. Oral itraconazole and a slow tapering dose of topical fluconazole over six weeks were continued for three months. The patient tolerated the treatment well and achieved complete recovery without any ocular complications (Figure 3).

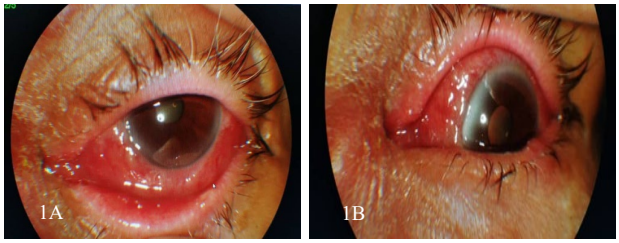

Figure 1a and 1b: Left eye bulbar granulomatous conjunctivitis

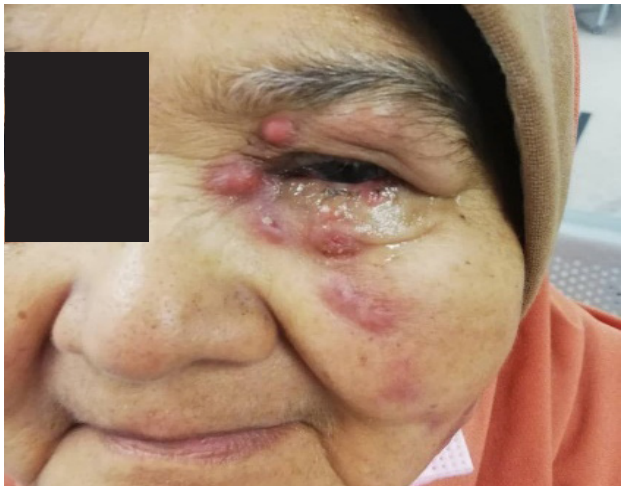

Figure 2: Multiple erythematous, granulomatous nodules over the left periorbital area extending to the left malar region

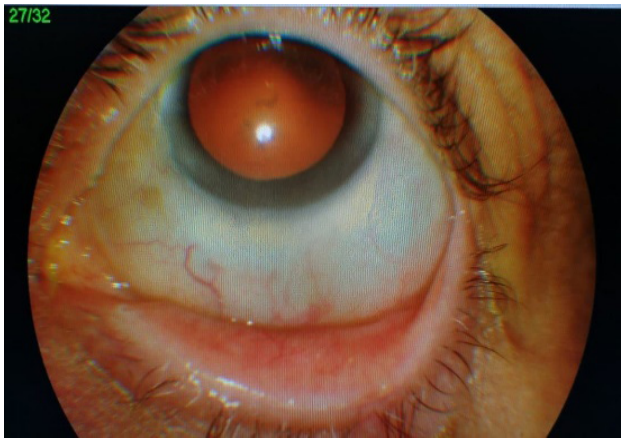

Figure 3: Complete resolution of the granulomatous conjunctivitis post-treatment

Discussion

Sporotrichosis is a subacute or chronic cutaneous mycosis caused by various species of *Sporothrix,* including *Sporothrix schenckii, S. brasiliensis, S. globosa, S. mexicana, S. albicans,* and *S.* lure. In Malaysia, *S. schenckii* is the primary pathogen responsible for feline sporotrichosis, while in Brazil, *S. brasiliensis* is the prevailing organism (3). The most common clinical manifestation of sporotrichosis is the lymphocutaneous form. Ocular involvement is rarer but can manifest as adnexal eye disease or intraocular manifestations such as anterior uveitis, endophthalmitis, and retinal granulomas (4). A literature search revealed eight English-language articles reporting cases of ocular sporotrichosis presenting with conjunctivitis (Table 1). The age of onset varied from three to 70 years, with no gender predilection. None of the individuals were immunocompromised, but all of them had prior contact with a cat or a history of being scratched by a cat. There were no reported cases of previous ocular trauma, which aligns with our presented case. The dimorphic fungus *S. schenckii* is commonly found in soil and decaying vegetation and typically infects humans through traumatic skin inoculation or, rarely, through inhalation, which may lead to hematogenous dissemination (1, 2). Although zoonotic transmission is rare, there is a growing trend of infections occurring after contact with cats that are contaminated with the fungus (4-6). These ailing cats can transmit the disease through bites or scratches. In our case, we believe that the fungus was transmitted non-traumatically from the ailing cat to our patient through direct contact with the cat's secretions. Scratching the lesion further facilitated the spread of the fungus, resulting in multiple lesions on the eye and skin.

Barros et al. (2) reported that ocular sporotrichosis primarily affects the ocular adnexa and can manifest as granulomatous conjunctivitis, Parinaud's oculoglandular syndrome (POS), dacryocystitis, or bulbar conjunctivitis. Granulomatous conjunctivitis, characterized by clusters of yellowish nodules on the bulbar and/or tarsal conjunctiva,

Table 1: Literature Review of Reported Cases of Ocular Sporotrichosis

No	Case report	Age	Sex	Comorbid	Contact with sick cat/ cat scratch	H/O Ocular trauma	Ocular lesion			Cutaneous lesion	Laterality of ocular involvement	Culture / PCR	Dosage of Oral Itraconazole	Treatment duration (months)	Ocular Sequelae	Complete recovery
							Conjunctivitis	Eyelid	LN							
1	Present	70	F	Nil	Yes	No	Tarsal and bulbar	Yes	Yes	Yes	Left	-ve	400 mg OD	3	No	Yes
2	Aidar et al. 2022 (5)	17	M	Nil	Yes	No	Tarsal	No	Yes	No	Left	-ve	200 mg OD	2	No	Yes
		6	M	Nil	Yes	No	Tarsal and bulbar	No	Yes	No	Right	No	100 mg BD	2	No	Yes
		18	F	Nil	Yes	No	Tarsal and bulbar	No	No	No	Right	No	200 mg OD	1	No	Yes
3	Liborio et al. 2021 (6)	40	F	Nil	Yes	No	Tarsal	Yes	Yes	No	Left	+ve	100 mg OD	2.5	No	Yes
4	Lemes et al. 2021 (7)	3	M	Nil	Yes	No	Tarsal	No	Yes	Yes	Right	+ve	NR	NR	No	Yes
		12	M	Nil	Yes	No	Tarsal and bulbar	Yes	No	Yes	Left	+ve	NR	NR	No	Yes
5	Gameiro et al. 2020 (8)	13	F	Nil	Yes	No	Tarsal	Yes	Yes	Yes	Left	+ve	200 mg OD	3	No	Yes
6	Yamagata et al. 2017 (9)	14	M	NR	NR	No	Tarsal	Yes	Yes	Yes	Right	+ve	200 mg OD	NR	No	Yes
		68	F	NR	NR	No	Tarsal and bulbar	No	No	Yes	Right	+ve	200 mg OD	9	Conjunctival fibrosis	Yes
		46	F	NR	NR	No	Tarsal	Yes	Yes	Yes	Left	+ve	200 mg OD x 2/12 then 400 mg OD x 6/12	8	symblepharon	Yes
7	Ling et al. 2018 (10)	18	F	Nil	Yes	No	Tarsal	No	No	No	Left	+ve	200 mg BD	6	No	Yes
8	Medeiros et al. 2016 (11)	59	F	Nil	Yes	No	Tarsal and bulbar	No	Yes	Yes	Left	+ve	200 mg OD	2	No	Yes
9	Ferreira et al. 2014 (4)	21	M	Nil	Yes	No	Tarsal	No	Yes	Yes	Right	+ve	100 mg OD	NR	NR	NR

M = Male; F = Female; LN = Lymph nodes, NR = not reported, +ve = positive, -ve = negative, BD = bis in die (twice daily), OD = omne in die (once daily)

accompanied by conjunctival hyperemia and mucopurulent discharge, is the most common presentation (3). POS is diagnosed when granulomatous conjunctivitis is associated with lymphadenopathy on the same side. Our review of the literature revealed similar findings; all patients presented with granulomatous conjunctivitis, with 64% of them having associated lymphocutaneous lesions. The preauricular, submandibular, and anterior cervical lymph nodes are commonly affected. In our case, the subacute course of granulomatous conjunctivitis with an unresponsive lymphocutaneous lesion raised strong suspicion of a fungal infection. The patient's history of contact with an ailing cat infected with sporotrichosis further supported the diagnosis of ocular and lymphocutaneous sporotrichosis. Intraocular involvement was not observed in our patient, which is consistent with Almeida-Paes et al. (1) findings that intraocular hemorrhage is rare and occurs because of hematogenous dissemination from multiple systemic lesions.

The treatment approach for ocular sporotrichosis is similar to that for the cutaneous form. Clinical suspicion is the key to diagnosis, and culture remains the gold standard (5). Conjunctival swabs or biopsies are commonly performed to culture and isolate the fungus for confirmation. Histopathological findings are nonspecific and can resemble other pyogenic processes and granulomas. Among the reported cases, 71% showed positive isolation of Sporothrix species. In our case, the conjunctival swab tested negative for *S. schenckii*, but a skin biopsy revealed granulomatous inflammation with mixed inflammatory cell infiltration. Although periodic acid Schiff staining did not detect fungal bodies, we made a presumed diagnosis of ocular and lymphocutaneous sporotrichosis based on the patient's history and clinical findings. The initiation of oral itraconazole (400 mg OD) led to a favourable response, and treatment was continued for three months. No side effects were observed during the course of treatment, and the patient achieved complete recovery without ocular complications. This outcome is consistent with our literature review, which indicated that 79% of patients were treated with oral itraconazole (200-400 mg/day) for one to nine months.

Out of the documented cases, two patients experienced symblepharon and conjunctival fibrosis, resulting in a delay in the initiation of systemic antifungal treatment. However, our patient received early treatment, which prevented any ocular complications. Based on the clinical correlation, we propose early administration of oral itraconazole for presumed ocular sporotrichosis, as it can be beneficial. Obtaining tissue samples for culture can be invasive, and the prolonged duration required for positive culture results may delay treatment. Therefore, prompt and appropriate treatment is crucial to completely resolve the lesion and prevent any ocular complications.

Conclusion

In conclusion, although ocular sporotrichosis is rare in Malaysia, it can still occur and present as granulomatous conjunctivitis. Unfortunately, it is often mistakenly treated as bacterial conjunctivitis, posing a high risk of ocular complications. Therefore, ophthalmologists should consider ocular sporotrichosis as part of the differential diagnosis for granulomatous conjunctivitis, particularly in patients with a history of contact with a sick cat or a cat scratch, along with associated lymphocutaneous lesions.

Conflicts of interest

There is no conflict of interest.

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Informed consent

Both verbal and written informed consent were acquired from the patient to include her participation in this case report.

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