

Case Series of Parinaud's Oculoglandular Syndrome Caused by Fungal Infection

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Abstract

We report 5 cases of fungal-related Parinaud's oculoglandular syndrome (POS), presenting to Hospital Sultanah Aminah from June 2017 to June 2019. This is a retrospective case series. Four patients presented with unilateral POS while another patient presented with unilateral granulomatous conjunctivitis which did not respond to a two-week course of oral doxycycline (4 patients) or a three-day course of oral azithromycin (1 patient). While all patients had contact with cats, only one had a history of being scratched by a cat. Three patient's pet cats were receiving treatment for 'Sporo' skin infection by their veterinarians while another two patients had no history suggestive of their cats having active infection. The first patient who presented with POS following her cat scratching her leg, had a skin biopsy done and received a course of antifungal antibiotics from a dermatologist when it was discovered that the cat had a fungal skin infection. Both her leg skin lesion and POS completely resolved. Conjunctival biopsies were performed in other 4 patients at 2 weeks to 1 month after their first presentation in view of poor response to antibiotics.

Two patients' conjunctival fungal culture and sensitivity showed a growth of *Sporothrix schenckii*. Both symptoms and conjunctival lesions resolved completely after taking a course of oral Itraconazole for 6 weeks (1 patient), 3 months (3 patients) and 4 months (1 patient). POS is most commonly caused by *Bartonella hensalae* infection related to a cat's scratch. Detailed history taking is important in diagnosing infection by other microorganisms such as fungi especially when the lesion does not respond to conventional antibiotic therapy. Besides clinical monitoring, conjunctival culture and sensitivity support the diagnosis.

Keywords: fungal infection, Parinaud's oculoglandular syndrome, sporotrichosis

Introduction

Parinaud's oculoglandular syndrome (POS) is a rare medical condition characterized by unilateral granulomatous conjunctivitis associated with ipsilateral cervical and either pre or retro auricular lymph adenopathy [1]. It has been associated with different etiologic agents, including bacteria,

viruses and fungi [2]. In fungal infections, the causative agents include sporotrichosis (*Sporothrix schenckii*), blastomycosis (*Blastomyces dermatitidis*), and coccidioidomycosis (*Coccidioides immitis*) [1].



Materials and methods

We conducted a retrospective review of the clinical records of 5 patients with clinical and microbiological evidence of fungal related POS presenting to Hospital Sultanah Aminah from June 2017 to June 2019.

Results

A total of 5 patients were diagnosed of fungal related POS within this study period.

The demographic features and other clinical information of these 5 fungal related

POS patients are summarized in **Table 1**. The mean age of presentation is 45 (range 13-62 years). 4 out of 5 patients were in the middle-aged and elderly populations (47-62 years). 3 out of 5 cases were female. 2 out of 5 patients had underlying diabetes mellitus and hypertension while the rest had no systemic illness.

Case	Age	Gender	Medical illness	History of presenting illness	Cat exposure	Physical examination
1	62	Male	Nil	Painless left eye redness with eye discharge for 2 weeks	Kept a pet cat undergoing treatment for a skin lesion called 'sporo"	<ul style="list-style-type: none"> • Left conjunctival granuloma over tarsal and bulbar conjunctiva • No lymphadenopathy
2	54	Female	DM, HPT	Painful left eye redness, with eyelid swelling for 3 weeks	Kept a pet cat receiving treatment for 'Sporo' skin infection by veterinarian	<ul style="list-style-type: none"> • Left conjunctival granuloma over tarsal and bulbar conjunctiva (Figure 1 and 2) • Enlarged left preauricular lymph node
3	13	Female	Nil	Painful right eye redness with eye discharge for 3 days	<ul style="list-style-type: none"> • Had exposure to many cats at home • No cat scratch or bite 	<ul style="list-style-type: none"> • Right conjunctival granuloma over tarsal and bulbar conjunctival • Enlarged right postauricular lymph node
4	47	Female	Nil	Left eye redness, swelling with itchiness for 1 week	<ul style="list-style-type: none"> • History of cat scratch over left leg 2 weeks prior to eye symptoms • The cat was receiving treatment for 'Sporo' skin infection 	<ul style="list-style-type: none"> • Left conjunctival granuloma over tarsal and bulbar conjunctiva • Enlarged left preauricular lymph node
5	50	Male	DM, HPT	Painful right eye redness and swelling for 2 weeks	<ul style="list-style-type: none"> • Had pet cats at home • No history of cat scratch or bite 	<ul style="list-style-type: none"> • Right conjunctival granuloma over tarsal and bulbar conjunctiva (Figure 3 and 4) • Enlarged right cervical lymph node

4 patients presented with acute unilateral POS while another patient presented with acute unilateral granulomatous conjunctivitis without lymphadenopathy. While all patients had contact with cats, only 1 had a history of being scratched by a cat. 3 patient's pet cats received treatment for 'Sporo' skin infection by their veterinarians while another 2 patients had no history suggestive of their cats having active infection. The first patient who presented with POS following her cat

scratching her leg, had a skin biopsy done by a dermatologist which showed no growth from the culture. In other 4 patients conjunctival biopsies were performed at 2 weeks to 1 month after their first presentation in view of poor response to antibiotics. 2 patients' conjunctival fungal culture showed a growth of *Sporothrix schenckii* while the other 2 patients' conjunctival fungal culture yielded no growth. Bartonella serology was negative in all 5 patients.

Figure 1: Granuloma over left lower tarsal and bulbar conjunctiva

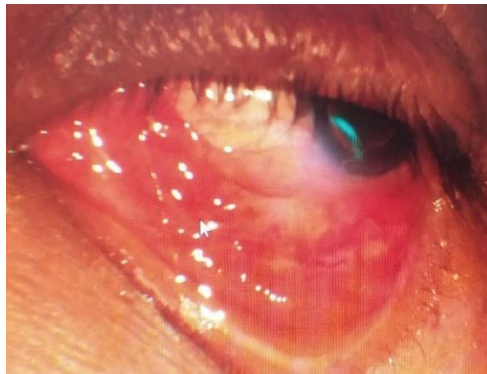


Figure 2: Complete resolution of left conjunctival granuloma



Figure 3: Granuloma over right upper tarsal and bulbar conjunctiva



Figure 4: Complete resolution of right conjunctival granuloma



Both symptoms and conjunctival lesions resolved completely after taking a course of oral Itraconazole for 6 weeks (1 patient), 3 months (3 patients) and 4 months (1 patient).

Discussion

The most common cause of Parinaud's oculoglandular syndrome (POS) has been cat scratch disease caused by *Bartonella henselae* infection. [1]. In this case series, 2 out of 5 cases were diagnosed of ocular sporotrichosis while 3 patients were clinically treated as fungal infection.

Sporotrichosis is an infection caused by fungi of the *Sporothrix* complex. It is a common plant saprophyte found in tropical and subtropical regions [3]. Sporotrichosis usually occurs by traumatic inoculation of soil, plants and organic matter contaminated with fungus [2]. Close interaction with cats plays a key role in transmission [2]. Cats constitute a reservoir of this agent as they have very close contact with contaminated soil and organic matter [2].

All 5 cases in this case series had exposure to cats. 1 patient received treatment for cutaneous sporotrichosis by dermatologist while 2 patients' pet cats were treated as cutaneous sporotrichosis by their veterinarians. The diagnosis of fungal infection in POS is usually confirmed by isolation of fungus in culture, as in 2 cases in this case series. Another 3 patients were treated as fungal infection despite of negative

finding in culture due to poor response to antibiotic. Treatment of fungal infection in POS is oral Itraconazole 100mg OD. Duration of antifungal varies depending on clinical condition, as in this case series, duration of antifungal ranges from 6 weeks to 20 weeks.

In conclusion, detailed history taking is important in diagnosing infection by other microorganisms such as fungi especially when the lesion does not respond to antibiotic therapy. Besides clinical monitoring, conjunctival culture supports the diagnosis.

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References

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