CASE REPORT

Medical management of suspected *Paecilomyces* sp. deep keratitis

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SUMMARY

A 74-year-old woman developed a deep keratitis presenting with an endothelial plaque without overlying stromal involvement. Owing to the characteristic clinical appearance, she was provisionally diagnosed with a *Paecilomyces* sp. keratitis. Corneal scrapings and aqueous humour samples were culture negative. The difficulties in diagnosing *Paecilomyces* sp. keratitis are discussed. The patient's keratitis was managed with a combination of topical, oral and intracameral injections of voriconazole. This medical management strategy was successful in achieving good infection control and visual outcome. We report a rarely encountered presentation of keratitis in which the clinical appearance and response to voriconazole are highly suggestive evidence of a *Paecilomyces* sp. keratitis. We believe the combination of topical, oral and intracameral injections of voriconazole represents a novel treatment approach for *Paecilomyces* sp. keratitis. Our case supports the potential for a medical approach as initial treatment in this otherwise devastating form of oculomycosis.

BACKGROUND

Paecilomyces is a genus of saprophytic, filamentous fungus that, although rare, is emerging as a significant cause of oculomycosis, particularly in Australia.¹ The consequences of *Paecilomyces* sp. keratitis are often devastating, resulting in endophthalmitis, perforation and enucleation.¹⁻⁴ Previously reported successes with medical therapy are limited to cases with superficial keratitis manifesting as a corneal ulcer.^{3, 5} We report a case of deep keratitis presenting with an endothelial plaque and clear stroma, presumed to be caused by *Paecilomyces* sp. This involved a novel combination of topical, oral and intracameral injections of voriconazole.

CASE PRESENTATION

A 74-year-old woman was referred to the Royal Brisbane and Women's Hospital, Brisbane, Australia, with pain, redness and decreased vision in her left eye. She had a history of a penetrating keratoplasty in this eye for Fuch's endothelial dystrophy 26 years prior, and a more recent phacemulsification and intraocular lens implantation 11 months earlier that improved her best spectacle-corrected visual acuity to L 20/100. The patient was otherwise systemically well with no history of immunosuppression, including no current topical steroid use. There was no history of trauma, contact lens wear or risk of occult penetrating eye injury. She denied exposure to fertilisers, pesticides, tank water or agricultural contamination.

On presentation, her best spectacle-corrected visual acuity was R 20/60 and L hand movements. The left conjunctiva was injected and there was a 2 mm×1.5 mm epithelial defect inferior to the visual axis of the graft, with a 2 mm hypopyon (figure 1). Centrally, there was an irregular endothelial plaque with clear overlying stroma (figure 2). The intraocular pressure was 11 mm Hg. Owing to the characteristic clinical appearance, the patient was provisionally diagnosed with a *Paecilomyces* sp. keratitis and admitted to hospital. Treatment was initiated with guttae voriconazole 1% one hourly and guttae atropine 1% twice daily. The hypopyon subsequently increased and she underwent anterior chamber paracentesis and a 0.1 mL intracameral injection of voriconazole 0.1%. The specimen of anterior chamber fluid containing the removed hypopyon was culture negative and thus represented a sterile inflammatory reaction rather than endophthalmitis. With institutional infectious diseases departmental approval, twice daily 0.1 mL intracameral injections of voriconazole 0.1% were added to the topical hourly regimen. This procedure was performed using an aseptic technique at the slit-lamp. After 4 days of treatment, the patient's visual acuity had improved to counting fingers, the endothelial plaque had reduced and there was no reaccumulation of the hypopyon. The epithelial defect persisted with no stromal infiltrate. The topical voriconazole 1% was reduced to hourly during waking hours and the twice daily intracameral injections were continued. After 7 days of this management, the patient was discharged on guttae voriconazole 1% hourly during waking hours.

Figure 1 Anterior segment photograph demonstrating hypopyon, central irregular endothelial plaque and small epithelial defect inferior to the visual axis occurring in a penetrating keratoplasty.

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Figure 2 Slit-lamp photograph demonstrating the irregular endothelial plaque and overlying clear corneal stroma.

OUTCOME AND FOLLOW-UP

At 1 week after discharge, the epithelial defect had resolved and the visual acuity was stable at counting fingers. There was no hypopyon, however, keratic precipitates had deposited over a large portion of the inferior cornea and there was an increase in the endothelial plaque formation. The patient was readmitted with resumption of twice daily 0.1 mL intracameral voriconazole 0.1%, started on oral voriconazole 200 mg twice daily and continued on guttae voriconazole 1% one hourly. Her anterior chamber reaction and endothelial plaque reduced and after 7 days, she was discharged on guttae voriconazole 1% one hourly during waking hours and oral voriconazole 200 mg twice daily with regular outpatient review. At 3 months follow-up, her best spectacle-corrected visual acuity had improved to 20/160. The irregular endothelial plaques had resolved, with residual corneal haze and pigmented keratic precipitates (figure 3). The guttae voriconazole 1% was reduced to six times a day and the oral voriconazole 200 mg twice daily continued for a total of 6 months.

Figure 3 Anterior segment photograph at 3 months follow-up showing resolution of the endothelial plaque, residual anterior stromal scarring and pigmented keratic precipitates.

DISCUSSION

We describe a case of presumed *Paecilomyces* sp. keratitis manifesting as an endothelial plaque without overlying stromal involvement. This uncommon infection often requires a combined medical and surgical approach to achieve resolution.^{4 6} Hirst *et al*⁴ described three cases of deep *Paecilomyces lilacinus* keratitis presenting, similarly to our patient, with an endothelial plaque, clear overlying stroma and no obvious source of fungal inoculation. Deep corneal biopsies were required to identify the organism in all cases, two of which were culture positive; however, the third case demonstrated suggestive histological features only. Two of these cases developed corneal perforation, one during deep corneal biopsy, and the other following corneal biopsy. All three cases required corneal grafting to control the infection. The authors proposed that patients presenting in this way might be examples of endogenous fungal keratitis that had not previously been described.

The diagnosis of *Paecilomyces* sp. keratitis is often difficult and delayed. Not uncommonly, corneal scrapings, corneal biopsies, ocular fluid samples and even excised corneal buttons, are culture negative.^{6 7} Characteristically, the stroma is clear as in our case, without a corneal infiltrate to scrape. The aqueous specimen including the thick hypopyon proved to be a sterile inflammation and not an endophthalmitis. Deep corneal biopsy would have been considered in the context of no clinical improvement with initial management, however, this increases the potential for conversion to an endophthalmitis. In the present case, the diagnosis of a *Paecilomyces* sp. keratitis can only be presumed as there was no definitive identification of the organism. The clinical appearance and the response to the voriconazole are, however, highly suggestive evidence.

Paecilomyces keratitis often follows a relentless course, commonly culminating in endophthalmitis, perforation and enucleation.^{1 3 4} Visual outcomes are often poor despite both medical and surgical management.¹ Deep infections such as the present case are associated with poorer outcomes. Other factors associated with poor outcomes include corticosteroid exposure and treatment with antifungals other than imidazoles.⁵ *Paecilomyces* sp. is highly resistant to conventional antifungal agents such as natamycin and amphotericin B, however, favourable outcomes have been reported with voriconazole.^{1 5 7 8} The few cases successfully managed medically share characteristics predictive of favourable outcomes mentioned above.^{3 5 9} We are not aware of another reported case of deep *Paecilomyces* sp. keratitis managed successfully without surgery and we believe our case supports the potential for a medical approach as initial treatment in this otherwise devastating form of keratitis.

Learning points

- ▶ *Paecilomyces keratitis* often results in devastating visual and ocular outcomes including enucleation.
- ▶ Confirmation of deep *Paecilomyces* infections is often difficult to obtain without corneal biopsy because scraping the overlying clear stroma will not sample the infected tissue.
- ▶ Corneal biopsy carries a risk of endophthalmitis and subsequent enucleation.
- ▶ Combined topical, intracameral and systemic voriconazole may provide a successful non-surgical management strategy with preservation of the eye and also vision.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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