A case of non-traumatic *Purpureocillium lilacinum* (*Paecilomyces lilacinus*) endophthalmitis in a child

Harrison Y. Bennett,a,b,* Shaheen P. Shah b

a University of Queensland, School of Medicine, Australia
b Queensland Children’s Hospital, 501 Stanley St, South Brisbane, QLD, 4101, Australia

**ARTICLE INFO**

**Keywords:** Fungal Endophthalmitis Purpureocillium Australia

**ABSTRACT**

**Purpose:** The aim of this report is to cover a novel presentation and subsequent management of *Purpureocillium* (*Paecilomyces*) oculomycosis in a child, and to review the available literature on *Purpureocillium* endophthalmitis.

**Observations:** This report is of a four year old boy from Australia. There have been 13 previous reports of *Purpureocillium* endophthalmitis, comprising 30 adult cases.

**Conclusions:** AND IMPORTANCE. *Purpureocillium* is an emerging ocular infection, associated with use of this fungus as a biological control agent. This case highlights the importance of early consideration of intraocular fluid sampling in a case of vitritis non-responsive to steroid treatment.

The first reported case of atraumatic *Purpureocillium lilacinum* endophthalmitis, occurring in a child. All published *Purpureocillium* endophthalmitis cases are reviewed.

1. Introduction

*Purpureocillium lilacinum* (formerly classified as *Paecilomyces lilacinus*) is a saprophytic filamentous fungus and opportunistic pathogen that can cause severe oculomycosis. 1 *P. lilacinum* is ubiquitous worldwide, but reports of severe ocular disease are primarily from Australia and in particular the state of Queensland. 1, 2, 3 We report the first case of *Purpureocillium* oculomycosis in a child, an endophthalmitis occurring in an immunocompetent four year old boy. This is of public health concern due to an absence of ocular disease, surgery, or significant trauma preceding infection.

2. Case report

A four year old Caucasian boy presented with a one week history of a red and painful right eye, that became increasingly painful in the two days prior to presentation. The boy was otherwise well; there were no systemic symptoms, no past medical history, no regular medications, and immunisations were up to date.

On examination, visual acuity was right eye ‘count fingers’, left eye 6/6 (Lea symbols). Intraocular pressures (IOP) were right eye 9 mmHg, left eye 11 mmHg (iCare). Anterior segment examination of the right eye demonstrated intense diffuse conjunctival injection, clear cornea with no epithelial defect and fine keratic precipitates, and 4+ anterior chamber cell with fibrin. Hazy posterior segment examination of the right eye demonstrated moderate vitritis, and a white lesion (suspected granuloma) extending from the optic nerve head with surrounding retinal oedema (Fig. 1A). Left eye was unremarkable.

Pending investigations, intensive topical prednisolone and cyclopiaegia were started. Chest X-ray, urinalysis, and all blood results (including eosinophils) were normal, apart from mildly elevated inflammatory markers. A hypopyon and dense vitreous inflammation developed, and oral prednisolone (1mg/kg) and oral azithromycin were commenced. Due to minimal response to treatment, deteriorating vision, and persistent ocular hypotension, vitrectomy was performed with aqueous and vitreous biopsy, and intravitreal administration of vancomycin and ceftazidime. Seven days post vitrectomy, *Purpureocillium* species was grown on vitreous culture. Panfungal PCR subsequently returned positive for *Purpureocillium lilacinum* DNA (ICPMR, Westmead, NSW Health). Topical and oral voriconazole were commenced and oral steroids weaned. Anterior segment inflammation gradually subsided. Endophthalmitis may have originated as a scleritis, as a nodular scleritis persisted as anterior segment inflammation subsided.

Voriconazole was ceased due to rash and hallucinations after six weeks, and oral posaconazole treatment commenced for a further two weeks. Lensectomy, vitrectomy, and silicone oil was performed at week...
seven for mature cataract and persistent ocular hypotension. Two months post cessation of treatment, the globe was maintained but with vision of ‘no perception of light’ and IOP of 3 mmHg.

3. Discussion

*Purpureocillium lilacinum* is an uncommon but devastating cause of oculomycosis. Presentation is typically with a keratitis, but *P. lilacinum* warrants special consideration in atypical uveitis, particularly in Australia. Vegetable matter trauma is a significant risk factor, and increasing reports of human disease have been concurrent with the adoption of *P. lilacinum* as a biological control agent for root-rot nematodes in agriculture. Infection often occurs in the setting of immunosuppression, and was classically described following ocular surgery or trauma. Recent reports from Queensland suggest a significant rate of atraumatic keratitis cases with intact epithelium.

Microbiological diagnosis of *P. lilacinum* infection is often delayed by weeks or months due to the non-specific and subacute clinical presentation. Diagnosis requires a high index of clinical suspicion, and often requires repeated sampling of intraocular fluid. Morphological diagnosis (Fig. 1) is difficult due to the fungus’ unique ability to sporulate adventitiously (unlike other filamentous fungi) and DNA sequencing of the internal transcribed spacer (ITS) region is recommended.

On review of the literature, there have been 13 previous reports of *P. lilacinum* endophthalmitis, comprising 30 adult cases (Appendix A – Table 1). 13 of these cases were secondary to contaminated intraocular lens irrigating solution in the USA in the 1970s. Two thirds had preceding ocular surgery, 5 had a history of ocular disease (scleritis or keratitis), and 3 had a history of trauma. 9 cases had a history of immunosuppression. Time to diagnosis ranged from nine days to many months, with the onset of infection sometimes difficult to distinguish from the ocular inflammatory disease that preceded it. 14 of 30 cases resulted in enucleation. A further 5 cases had very poor final visual acuity (‘counting fingers’ or worse). As in this report, all reported endophthalmitis cases underwent surgical management.

*P. lilacinum* is difficult to treat, with resistance to most available antifungal agents including the frequently used ocular antifungal agents natamycin and amphotericin B. Disease prognosis has improved with the availability of the new triazole antifungal agents voriconazole and posaconazole. Voriconazole may be delivered topically (with minimal toxicity and good epithelial penetration) and intraocularly, and therapeutic intraocular concentrations may be achieved with oral or intravenous use. Terbinafine has been described to have synergistic effect with voriconazole against *P. lilacinum*. Despite the availability of effective treatment, therapy must be prolonged (often over many months) and late stage relapses are common.

4. Conclusions

This case is of public health concern, reporting on the first case of *Purpureocillium lilacinum* endophthalmitis in an otherwise well child. In regions such as Australia with increased prevalence of *Purpureocillium* infection clinical suspicion must be high, with early consideration of intraocular sampling in cases of atypical inflammatory disease.

Patient consent

Approval was obtained from the Human Research Ethics Committee – Centre for Children’s Health Research Queensland. Consent was obtained from the patient’s legal guardians and was
documented in the electronic medical record.

**Funding**

No funding or grant support.

**Authorship**

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

**Declaration of competing interest**

The following authors have no financial disclosures: HB, SS.

**Acknowledgements**

Dr Sarah Kidd, National Mycology Reference Centre, SA Pathology.

### Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.ajoc.2022.101375.

### References

1. Luangsa-Ard J, Houbraken J, Van Doorn T, et al. Purpureocillium, a new genus for the medically important Paecilomyces lilacinus. *FEMS (Fed Eur Microbiol Soc) Microbiol Lett.* 2011;321(2):141–149.
2. Hirst LW, Choong K, Playford EG. Nontraumatic *Paecilomyces* Anterior Segment Infection. 2014;33(10):1031–1037.
3. Turner LD, Conrad D. Retrospective case-series of Paecilomyces lilacinus ocular mycoses in Queensland, Australia. *BMC Res Notes.* 2015;8:627.
4. Antas PRZ, Brito MMS, E Peixoto, Ponte CGG, Borba CM. Neglected and emerging fungal infections: review of hyalohyphomycosis by Paecilomyces lilacinus focusing in disease burden. *in vitro antifungal susceptibility and management.* 2012;14(1):1–8.
5. Chew R, Dorman A, Woods ML. Purpureocillium lilacinum keratitis: a case series and review of the literature. *Can J Ophthalmol.* 2016;51(5):382–385.