

Case Report

Delayed Bacterial Endotheliitis and Endophthalmitis 11 Years after Cataract Surgery

Elisabeth Poon^a Alexander Poon^b Penelope McKelvie^c Lewis Levitz^d
Ehud Zamir^e

^aMedical School, Monash University, Melbourne, VIC, Australia; ^bCorneal Unit, Royal Victorian Eye and Ear Hospital, Melbourne, VIC, Australia; ^cDepartment of Anatomical Pathology, St Vincent's Hospital, Melbourne, VIC, Australia; ^dCataract surgery, Vision Eye Institute, Melbourne, VIC, Australia; ^eCentre for Eye Research Australia, The Royal Victorian Eye and Ear Hospital, Melbourne, VIC, Australia

Keywords

Cataract · Cataract surgery · Endophthalmitis · Infectious disease · Inflammation/uveitis

Abstract

Infective endophthalmitis is an uncommon complication following intraocular surgery. Chronic endophthalmitis may present some time after intraocular surgery, making the diagnosis challenging. *Cutibacterium acnes* is a well-recognised causative agent of these chronic infections. Practitioners should be aware of the conditions required to culture this slow-growing organism. We report a case of delayed low-grade endophthalmitis presenting 11 years after cataract surgery. *Cutibacterium acnes* and *Staphylococcus warneri* were cultured from Descemet's membrane biopsy following three failed previous attempts at microbiological studies. Clinical features of the infection included discrete white granules on the iris, endothelium, and within the capsular bag of the patient's right eye. The patient presented with no signs of systemic infection and the left eye was normal on examination. Bullous keratopathy, secondary to endothelial dysfunction was a feature of this infection. This retrospective case report illustrates the prolonged periods for which *Cutibacterium acnes* can remain latent before causing clinical signs. While uncommon, endothelial involvement may occur and clinicians should consider low-grade infective endophthalmitis in cases with corneal oedema.

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Correspondence to:
Alexander Poon, alexpoon@ozemail.com.au

Introduction

Endophthalmitis following intraocular surgery accounts for 90% of endophthalmitis cases [1]. The most common presentation is of abrupt onset typically a week or less post-surgery. Symptoms on presentation may include pain and worsening of vision, while examination can reveal hypopyon and vitritis. Low-grade chronic endophthalmitis may present some time postoperatively without these signs and symptoms, making it difficult to diagnose correctly. It is the organisms of lower virulence, such as *Cutibacterium* (formerly *Propionibacterium*) *acnes* which are associated with this chronic and indolent process. A consecutive case series of delayed onset endophthalmitis post-cataract surgery found the mean time between surgery and diagnosis to be 343 days (range: 48–1,840, SD: 379) [2]. The longest reported delay in presentation of infectious endophthalmitis after intraocular surgery is 21 years [3]. We report a case of delayed postoperative low-grade endophthalmitis presenting 11 years after cataract surgery, with corneal endothelial involvement. Corneal endothelial dysfunction and bullous keratopathy have not been previously reported as features of *C. acnes* endophthalmitis. The CARE Checklist has been completed by the authors for this case report, attached as online supplementary material (for all online suppl. material, see <https://doi.org/10.1159/000531501>).

Case Presentation

An 88-year-old man presented with a 2-month history of painless blurry vision in the right eye. He had a history of cataract surgery and intraocular lens implantation 11 years earlier. His best-corrected visual acuity (BCVA) was 20/100 in the right eye and 20/25 in the left eye. There was sectorial corneal stromal oedema and bullous keratopathy in the inferior cornea. The endothelium and iris were diffusely speckled with chalky-white granules which also formed discrete clusters and slender linear deposits on both surfaces of the intraocular lens (Fig. 1, 2). The vitreous and fundus were normal.

A tentative diagnosis of delayed postoperative endophthalmitis with *Cutibacterium acnes* was made and an anterior chamber paracentesis was performed. Cultures and gram stain microscopy were negative after 2 weeks of incubation. Polymerase chain reaction was requested but not available at our institution. Dexamethasone drops were administered every 2 hours, resulting in a visual improvement to 20/40; however, the inferior corneal oedema increased. Empirical treatment with oral moxifloxacin, 400 mg daily for presumed *C. acnes* endophthalmitis was commenced. Specular microscopy and confocal microscopy showed a reduced central cell density count of 955/mm², and discrete, refractile endothelial deposits were seen (Fig. 3). Removal of the intraocular lens and the capsular bag was recommended but the patient declined.

Two months after his presentation, a right anterior chamber and capsular bag washout/biopsy were performed, for microbiologic and histopathologic studies. A chalky “cast” was removed from within the capsular bag, and an adherent film, containing the linear white deposits, was peeled off the anterior IOL surface. Iris deposits were also removed for analysis. Intravitreal vancomycin 2% and intracameral moxifloxacin 0.17% were injected. The cultures were negative after 2 weeks of incubation, but the histopathologic examination was equivocal, revealing calcified granular debris but no organisms. Gram stain of the debris was reported as being negative.

The cornea was clear when examined on postoperative day 3, but on day 5 became diffusely oedematous with stromal and microcystic oedema. The oedema continued to worsen over the following 3 weeks. Three weeks postoperatively, a Descemet’s membrane detachment was noted, with worsening bullous keratopathy and visual acuity of less than 20/200. Sulphur hexafluoride tamponade of Descemet’s detachment was performed. Anterior

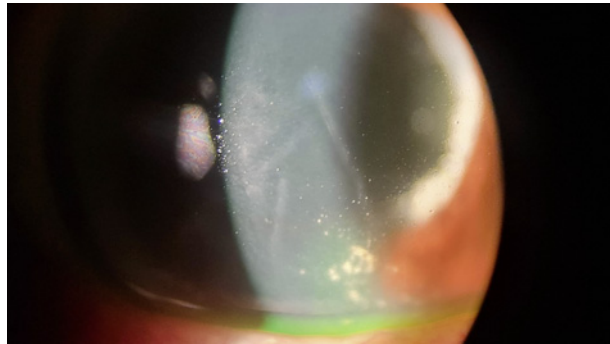


Fig. 1. Endothelial granules before wash out.

chamber aspirate was again sent for microbiological examination and culture. No organisms were seen or cultured after 2 weeks of incubation. By 6 weeks, the BCVA improved to 20/20. Two months later, the cornea was again oedematous, showing signs of endothelial failure. Descemet's membrane endothelial keratoplasty was performed. The Descemet's membrane was sent for histological and microbiological examination. *C. acnes* and *Staphylococcus warneri* were cultured after 12 days. Histological examination demonstrated very few endothelial cells and no evidence of inflammation or organisms on special stains.

Ten weeks after Descemet's membrane endothelial keratoplasty, approximately 1 year after the initial presentation, the BCVA was 20/25 and the cornea was clear, with no deposits but there were residual white chalky deposits on the iris and pupil margin as well as on the posterior capsule as seen in Figures 4 and 5. A vitreoretinal opinion was sought and the patient again decided to leave the intraocular lens and posterior capsule in situ as its removal would most likely lead to visual deterioration. Removal of the intraocular lens and posterior capsule would be considered if there was a progression of intraocular inflammation and worsening vision.

Discussion

Coagulase-negative streptococci account for 70% of culture-positive post-cataract endophthalmitis cases [4]. *C. acnes* is a recognised cause of postoperative granulomatous uveitis following extracapsular implant surgery [5]. It is an anaerobic gram-positive bacillus that is found ubiquitously on the skin. Known to cause device-related infections, it is thought to create a biofilm around the prosthesis and be difficult to eradicate medically [6]. In the eye, the organism is sequestered in lens remnants and capsule. *S. warneri*-associated endophthalmitis is rarely documented but has been found alone and concomitantly with other organisms [6, 7]. It is a coagulase-negative staphylococcus which is commonly found as a skin commensal and therefore may be a contaminant in this case. However, it has also been associated with bacteraemia, urinary tract infections, and mastitis [8].

Three initial attempts at culture and pathology failed to conclusively reveal the presence of any microorganism. All three cultures were incubated for 2 weeks before being declared negative as per the protocol for suspected *C. acnes* at our institution. Finally, histological and microbiological examination of the Descemet's membrane specimen cultured in enrichment broth revealed the 2 organisms after 12 days. Despite negative studies, repeated sampling when there is a high index of suspicion is highly recommended. As shown by our three negative cultures, enough time should be allowed for *C. acnes* culture growth, which may take longer than the standard 14 day incubation period. Polymerase chain reaction has been shown to have a high rate of positive identification of the causative bacteria in chronic,

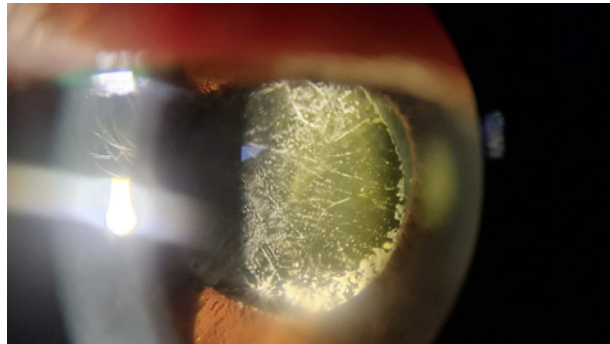


Fig. 2. IOL granules before wash out.

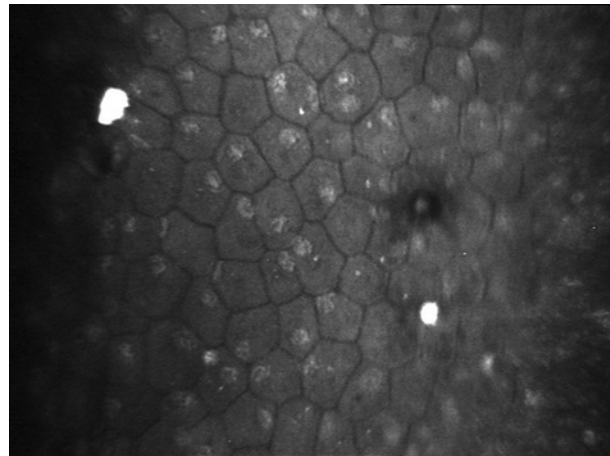


Fig. 3. Specular microscopy showing refractile granules on the endothelium.



Fig. 4. Clear cornea with iris and pupil margin deposits.

delayed onset endophthalmitis (84%) when compared to culture or microscopy of aqueous humour (0%) [9]. However, while this diagnostic tool has clear benefits, it is not readily available at all institutions.

Clues to the presence of *C. acnes* include white intracapsular plaques and chronic inflammation. While a previous case reported a single large endothelial deposit associated with

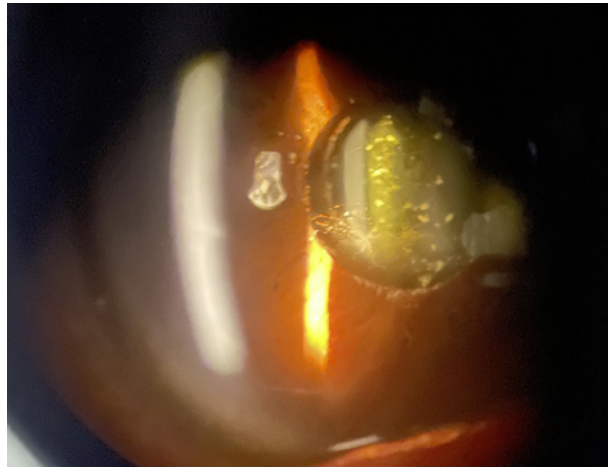


Fig. 5. Vitreous in the pupil plane and white deposits on the posterior capsule.

C. acnes endophthalmitis, endothelial involvement is uncommon [5]. Our case presented with significant endothelial involvement, unlike the case described by Abrahams. The negative histopathological examination and positive cultures of the Descemet's membrane may be explained by the lower sensitivity of histologic exams when compared to culture. Even in cases of fulminant acute endophthalmitis, bacteria are often not easily found in histological sections.

Our patient had cataract surgery performed 11 years previously, after which the patient had good vision. There was no known causative event which may have precipitated the inferior corneal oedema noted on presentation. The area of bullous keratopathy was directly above a relatively dense cluster of deposits on the endothelium, which led to our conclusion that the endothelial dysfunction was directly related to the infectious process. Furthermore, the specular microscopy did not show widespread endothelial loss as would be expected in pseudophakic bullous keratopathy. The oedema worsened after anterior chamber washout, but this worsening was not immediate as would be expected if it was purely related to intraocular manoeuvres. While it is possible that the intraocular manoeuvres played a role in this corneal oedema, the fact that bullous keratopathy was seen at presentation, and at the site of most endothelial deposits, indicates the primary insult was infectious rather than surgical.

Treatment of chronic *C. acnes* endophthalmitis usually involves a combination of intraocular antibiotics as well as surgery. In a literature review conducted by Fowler et al. [10], it was found that intraocular lens removal was the definitive treatment with 100% of cases reporting no recurrent inflammation, while 72% of cases who had intraocular antibiotics alone had recurrent inflammation. To date, our 88-year-old patient has been resistant to intraocular lens removal and capsulectomy due to his age, slow progression of disease, and good vision with a clear cornea.

Conclusion

C. acnes may remain latent for many years after cataract surgery before presenting as chronic low-grade endophthalmitis. Clinicians should be aware that the endothelium may be a site of infection and should be monitored to prevent endothelial failure. A positive culture may be difficult to obtain; however, cultures should be kept for longer than 14 days when suspicious of *C. acnes*.

Statement of Ethics

Ethical approval is not required for this study in accordance with local or national guidelines. Written informed consent for publication was obtained from the patient for publication of this case report and all accompanying images.

Conflict of Interest Statement

The authors declare that there are no conflicts of interest.

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Author Contributions

Elisabeth Poon collated information from all practitioners and prepared the manuscript. Alexander Poon, Ehud Zamir, Penelope McKelvie, and Lewis Levitz were all involved in the care of the patient presented in this case report and helped edit the manuscript.

Data Availability Statement

All data generated and analysed in this study were included within this article. Further enquires can be directed to the corresponding author.

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