Case Report

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Burkholderia cepacia keratitis with endophthalmitis

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Keratitis with endophthalmitis of the right eye occurred in a 78-year-old female following a complicated cataract surgery. Prompt intravitreal vancomycin and ceftazidime with topical fortified tobramycin and cefazolin treatments were started. The corneal, aqueous and vitreous cultures grew a *Burkholderia cepacia* complex (Bcc) strain on the fourth day. Restriction fragment length polymorphism analysis of the *recA* amplicon revealed *B. cepacia* genomovar I. The organism was found to be susceptible to ceftazidime, ciprofloxacin and ofloxacin. Topical ciprofloxacin was given immediately. At day 10, the pain relieved and the clinical condition of the patient improved with resolution of the purulent discharge, severe circumcorneal congestion and chemosis. The size of the corneal abscess and anterior chamber exudation decreased. The Bcc should be included among the bacterial species that may cause keratitis following intraocular surgeries.

Introduction

Burkholderia cepacia complex (Bcc) isolates are well-known Gram-negative, oxidase-positive, non-fermentative bacilli. They may cause severe infection in immunocompromised patients and in healthy individuals. The Bcc has been shown to be resistant to antiseptics such as benzalkonium chloride and chlorhexidine at standard or high concentrations (Middleton *et al.*, 2005). Ocular infections including keratitis and endophthalmitis are very rare in the literature (Del Piero *et al.*, 1985; Pathengay *et al.*, 2005; Eser *et al.*, 2006; Ying-Cheng *et al.*, 2006). This report describes what we believe to be the first case of Bcc keratitis with endophthalmitis following cataract surgery.

Case report

A 78-year-old female was admitted to us with decreased vision and severe pain in the right eye following a cataract operation. She was put on hourly topical lomefloxacin and dexamethasone eye drops. A stromal infiltrate with oedema around the clear corneal incision was noted on the first postoperative day by the operating surgeon as per records. Her visual acuity was no light perception in the right eye and 20/25 in the left eye on admission. Slit lamp biomicroscopy of the right eye revealed a purulent discharge, severe circumcorneal congestion, chemosis and almost a total corneal abscess. There was a 4.0 × 5.0 mm central epithelial defect overlying the dense abscess. B-scan ultrasonography revealed medium echo spikes throughout the vitreous cavity showing vitreous opacities without retinal detachment. Since the patient was diagnosed as having keratitis with acute postoperative endophthalmitis, immediate corneal scraping with aqueous and vitreous taps

were performed. She was given intravitreal injections of vancomycin (1 mg per 0.1 ml) and ceftazidime (2.25 mg per 0.1 ml). Hourly topical fortified tobramycin and cefazolin were added to the treatment.

Specimens were inoculated onto MacConkey agar, blood agar, Sabouraud medium, Eosin Methylene Blue agar and chocolate agar and into thioglycollate broth. Culture grew non-lactose-fermenting, mucoid, smooth colonies with a diameter of approximately 2 mm. The colonies were made up of Gram-negative rods. The Gram-negative rods were motile, catalase- and oxidase-positive, and nitrate reduction test-positive. The rods oxidized glucose, lactose and maltose. They were positive for lysine decarboxylation and negative for arginine hydrolysis. They grew well at 42 °C and were motile at 37 °C. The micro-organism was identified as a member of the Bcc by a BBL Crystal Enteric/Nonfermenter ID kit (Becton Dickinson). Subculturing the material using *B. cepacia* selective agar (BCSA) yielded the same organism on the fourth day.

In addition to laboratory diagnosis, we investigated the identity of the genomovar after culturing the organism. Cultures of the bacterial strains were suspended in 1 ml 10 mM Tris/HCl buffer containing 1 mM EDTA (TE buffer) and centrifuged at 10 000 *g* for 10 min. Supernatants were removed, and the resulting pellets were resuspended in 0.2 ml TE buffer. Genomic DNA was prepared using a high-purity PCR template kit (Roche Diagnostics). The Bcc *recA* gene was amplified using primers BCR1 and BCR2. For restriction fragment length polymorphism analysis, the Bcc *recA* amplicons were digested with *Hae*III and *Mn*II restriction endonucleases. Amplicons (10 µl) were added to the endonuclease along with the appropriate enzyme buffer, in accordance with the manufacturer's instructions, before incubation at 37 °C

Abbreviation: Bcc, *Burkholderia cepacia* complex.

for 2 h. PCR-amplified products were routinely analysed by electrophoresis in 2 % (w/v) agarose gels containing 40 mM Tris buffer and 20 mM acetate. Resolved DNA products were stained with ethidium bromide and viewed under UV light. Using primers BCR1 and BCR2, the *recA* gene product was successfully amplified from the genomic DNA of the organism. The resulting 492 bp amplicon was visualized after gel electrophoresis and staining with ethidium bromide. The isolate was identified as *B. cepacia* genomovar I.

The bacterium was found to be susceptible to ceftazidime, ciprofloxacin and ofloxacin, and was resistant to ceftriaxone, tobramycin and amikacin. Topical tobramycin (1.5 %) and cefazolin (5 %) were replaced with topical ciprofloxacin (0.3 %) as a result of the sensitivity studies. At day 10, the pain relieved and the clinical condition of the patient improved with resolution of the purulent discharge, severe circumcorneal congestion and chemosis. The patient was lost to follow-up after 1 month and the long-term status of the eye could not be documented.

Discussion

Bacterial endophthalmitis is one of the most feared complications of cataract surgery and may result in severe vision loss. Using standard methods for identification and isolation of both bacteria and fungi, coagulase-negative staphylococci account for nearly 60 % of all cases, and *Staphylococcus aureus* accounts for another 20 % of the total (Baron *et al.*, 1994; Collee *et al.*, 1996). The Bcc is known to be a rare cause of acute postoperative endophthalmitis (Del Piero *et al.*, 1985; Pathengay *et al.*, 2005; Eser *et al.*, 2006). To our knowledge, this is the first case of Bcc keratitis with endophthalmitis following a cataract surgery.

To date, four cases of postoperative endophthalmitis due to Bcc have been reported. The patient in the first case in the literature presented with chronic anterior uveitis following a combined glaucoma and cataract surgery and subsequently developed endophthalmitis (Del Piero *et al.*, 1985). Recurrent endophthalmitis caused by Bcc was an acute onset inflammation in an eye previously operated on for a cataract (Pathengay *et al.*, 2005). Eser *et al.* (2006) reported two endophthalmitis cases following uncomplicated cataract surgeries which improved with early vitrectomies. There is only one report of keratitis due to Bcc, which was treated successfully with topical ciprofloxacin in a patient who wore orthokeratology lenses (Ying-Cheng *et al.*, 2006). In our case, we assume that the clear corneal wound infection led to a severely infected cornea with endophthalmitis. However, the original source of the *B. cepacia* strain associated with this isolated case remains unknown, as it was not part of a nosocomial outbreak. Environmental water or dust may be possible sources, as these operations are now outpatient surgeries and the patients use eye shields for only a few hours following surgery.

Members of the Bcc have a natural resistance even to most of the powerful antibiotics that we use to treat infections in clinical practice. This multidrug resistance may be due to a drug efflux system and inducible chromosomal β -lactamases (Aaron *et al.*, 2000; Wigfield *et al.*, 2002). This multiresistance makes treatment of Bcc a problem. Drug sensitivity testing has shown that the Bcc is resistant to most antibiotics, including ceftazidime, ceftriaxone, tobramycin, amikacin and gentamicin, and sensitive to ceftazidime, ciprofloxacin, ofloxacin, piperacillin and imipenem in previous reports. Intravitreal vancomycin and ceftazidime were administered and topical fortified tobramycin and hourly cefazolin were started on admission. However, the organism was found to be sensitive to ciprofloxacin, and in 1 week, the pain relieved with improvement in the clinical condition of the patient.

All *Burkholderia* species possess very large genomes ranging between 6 and 9 Mb in size, and it is this huge genetic capacity which underpins their versatility in disease and natural biology. We suppose that due to the difficulties in accurately identifying this organism, the Bcc may be responsible for a larger number of human ocular infections than recognized. It should be included in the list of bacterial species that may cause postoperative infections.

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