

Burkholderia cenocepacia keratitis

Geeta Behera, Radha Sugumar¹, Sujata Sistla¹,
Mary Stephen, Subashini Kaliaperumal,
K Ramesh Babu¹

Case report: A 33-year-old lady with history of failed keratoplasty for decompensated cornea due to childhood trauma and secondary glaucoma, post glaucoma drainage implant, with pseudophakia in the right eye, developed bacterial keratitis following foreign body trauma to corneal graft. Corneal cultures yielded *Burkholderia cenocepacia* identified by matrix-assisted laser desorption ionization-time of flight mass spectrometry (MALDI-TOF- MS, bioMerieux, France). She healed with topical antibiotics (moxifloxacin 0.5%) in 1 month. Ours is the first report of ocular *Burkholderia cenocepacia* infection, possibly an under reported, aerobic, organism.

Key words: *Burkholderia cenocepacia*, keratitis, MALDI-TOF- MS

Ocular infection due to *Burkholderia spp* is rarely reported, most often in patients on topical steroids. There are many

species of *Burkholderia* that are found in nature, but most infections reported are by *Burkholderia cepacia*.^[1-6] Keratitis due to *Burkholderia cenocepacia* has never been reported so far.

Case Report

A 33-year-old lady presented with redness, watering, pain, and discharge in her right eye (RE) following foreign body trauma of 1 week. The trauma was with cement mix at a construction site. Her vision in RE was hand movements (HM) with accurate projection of light. The RE had prior diagnosis of failed keratoplasty for decompensated cornea due to childhood trauma and secondary glaucoma, post glaucoma drainage implant (AADI; Aurolab, Madurai, India), and pseudophakia. The RE had poor vision (HM), prior to trauma, and the patient was on long-term topical steroids. On examination, she had a central ulcer of size 4.1 mm × 2.6 mm in a slightly superior part of the corneal graft with surrounding infiltrate extending to 12 o'clock suture, forming an abscess, with diffuse graft edema [Fig. 1a and b]. The graft host junction was well apposed and sutures intact. There was no hypopyon and anterior chamber reaction could not be determined. She was advised eyedrops moxifloxacin 0.5% hourly round the clock, pending microbiological evaluation.

Corneal scrapings were inoculated on 5% sheep blood agar and MacConkey medium, which grew gray moist and nonlactose fermenting colonies, respectively, after 24 h of aerobic incubation. Gram staining showed slender gram-negative bacilli, motile, and oxidase-positive [Fig. 2a and b]. It was identified as *Burkholderia cenocepacia* by matrix-assisted laser

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Departments of Ophthalmology and ¹Microbiology, Jawaharlal Institute of Post-Graduate Medical Education and Research (JIPMER), Puducherry, India

Correspondence to: Dr. Geeta Behera, Jawaharlal Institute of Post-Graduate Medical Education and Research (JIPMER), Puducherry - 605 006, India. E-mail: geeta.anandaraja@gmail.com

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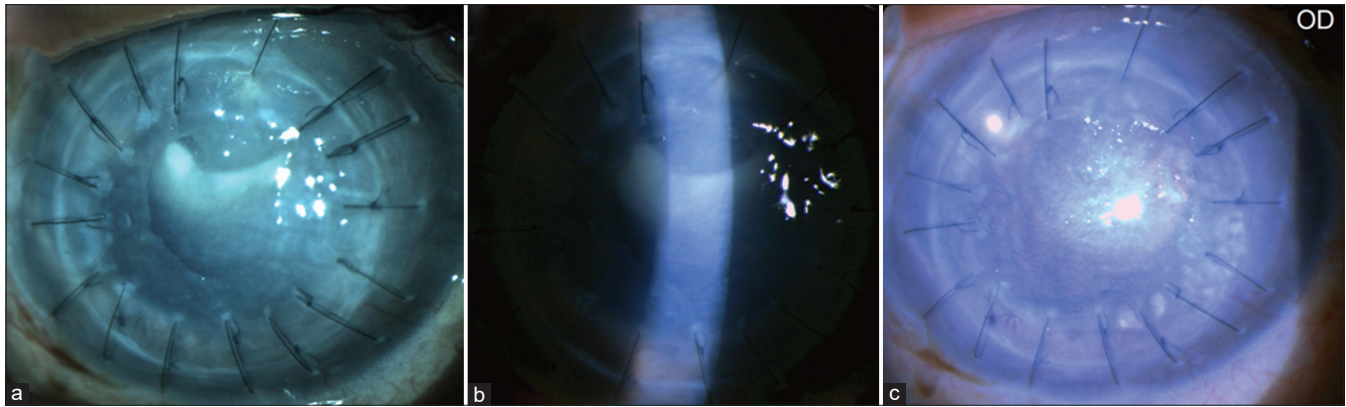


Figure 1: Slit-lamp photographs. (a) Diffuse photograph of graft ulcer at 12 o'clock with infiltrate and conjunctival congestion. (b) Slit photograph of the graft ulcer. (c) Complete healing of graft ulcer after 1 month with scarring

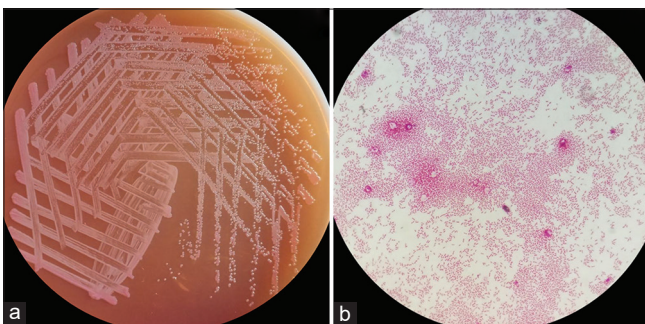


Figure 2: (a) MacConkey medium showing round nonlactose fermenting colonies of *B. cenocepacia*. (b) Gram staining performed from the colony showing gram-negative bacilli with parallel sides and round ends

desorption ionization-time of flight mass spectrometry system (MALDI-TOF-MS, bioMerieux, France) in our laboratory.^[7] In vitro susceptibility was carried out using VITEK-2 (bioMerieux, France). The organism was found susceptible to ceftazidime, ciprofloxacin, meropenem, minocycline, and cotrimoxazole. The VITEK-2 AST card does not have moxifloxacin and the susceptibility of the organism to ciprofloxacin cannot be extrapolated for moxifloxacin. As there are no CLSI breakpoints for moxifloxacin against *Burkholderia cepacia* complex (BCC), the susceptibility to this drug cannot be reported. However, it can be used for treatment. The ulcer decreased to less than half its original size with partial resolution of infiltrates after 1 week of treatment. So, treatment was continued and tapered to two hourly after 2 weeks and three hourly after 3 weeks. The ulcer healed completely after 1 month [Fig. 1c]. Her vision in RE remained HM with accurate projection of light.

Discussion

Burkholderia cepacia complex (BCC) is a ubiquitous group of oxidase producing, lactose nonfermenting, gram-negative bacteria composed of 24 closely related species as of now, but due to rapid mutagenic capability of the organism, many more species may emerge.^[8] Majority of keratitis cases reported have been due to *Burkholderia cepacia*.^[1-6] This organism has an intrinsic capacity to survive in various pharmaceutical products causing nosocomial outbreaks attributed to its inherent resistance to antibiotics and antiseptics. Other virulence factors include complex metabolic pathways, survival at a wide range of temperatures from 18 to 42°C,

biofilm formation, etc., which allow survival in various adverse environmental conditions.^[8] Therefore, even a single case should alert the clinician to take prompt action to prevent spread of infection. Phenotypic identification of BCC members is a tedious procedure and, for most, biochemical distinctions not possible.^[9] Among automated systems, MALDI-TOF-MS plays an important role in identifying BCC members. It works on the principle of mass spectrometry where identification is made by comparing peptide mass fingerprint (PMF) of unknown organisms with PMF in the system database, with studies showing good concordance to sequencing.^[7] Identification of organisms is very rapid (within 20 min), which aids in instituting appropriate antimicrobial therapy.

Our patient reported trauma and contamination with infected soiled water from cement-mix at a construction site. Additionally, she had been using topical steroids on a long-term basis, which compromised immunity of the ocular surface. Pharmaceutical contamination could be a possible source of infection, though we did not culture the steroid eyedrop that she was using. Had we done so and isolated the bacteria, it may have been identified as a possible source of the infection. In our case, it is more likely that the foreign body trauma was the source of infection due to a clear temporal association of trauma and onset of symptoms over the following week. There are few reports of keratitis due to BCC following trauma.^[4,6] Chaurasia *et al.* reported four cases of keratitis with BCC, of whom two cases were post-trauma, and one was on topical steroids.^[4] Keratitis has also been reported with contact lens use, following cataract surgery and LASIK.^[2,3,5,6] In immunocompetent individuals, it has been reported as chronic, indolent ulcers.^[10] Most reported good healing following appropriate identification and treatment. Unlike most other species of BCC, the strain we cultured was susceptible to most drugs with clinical improvement on moxifloxacin 0.5% eyedrops.

There are few cases of infectious keratitis reported with *Burkholderia spp.*, and it was first reported by Levy *et al.*, in a postkeratoplasty patient using contact lenses in 1985.^[1] This was prior to its classification as *Burkholderia spp.*, and it was reported as *Pseudomonas cepacia*. Before MALDI era, the *Burkholderia* genus itself was misidentified because oxidase positive nonfermenters were considered to be *Pseudomonas* species by most microbiologists. Conventional methods of *Burkholderia* species identification are laborious and usually not undertaken. This could be the possible reason for underreporting of cases.

Conclusion

Burkholderia spp should be considered in keratitis, especially in patients whose ocular surface immunity may be compromised due to topical steroids. Ours is likely the first report of *Burkholderia cenocepacia* keratitis.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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