Endogenous endophthalmitis secondary to *Burkholderia cepacia*: A rare presentation

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Burkholderia cepacia (previously known as *Pseudomonas cepacia*) is low virulent, gram negative bacilli, known to cause infections in immunocompromised hosts. There are reports about this organism causing keratitis, acute or delayed postoperative, or post traumatic endophthalmitis. Persistence of infection and poor visual outcome are well known complications of infection caused by this organism. Endogenous endophthalmitis due to *Burkholderia cepacia* is rare. There is no such case report available of endogenous endophthalmitis caused by these bacteria in the literature, where it is presented as retinal abscess and retinal vasculitis. Our aim is to report such a rare case from our hospital, which was treated with systemic and intravitreal antibiotics, with control of infection.

Key words: *Burkholderia cepacia,* endogenous endophthalmitis, retinal abscess

Access this article online

Quick Response Code:	Website: www.ijo.in
	DOI: 10.4103/ijo.IJO_1648_19

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Received: 16-Oct-2019 Revision: 09-Apr-2020
Accepted: 10-May-2020 Published: 23-Sep-2020

Endogenous endophthalmitis is rare and accounts for approximately 2-15% of all cases of endophthalmitis[1] and occurs when infectious organisms are spread from distant focus to the eye through the blood stream.[2] In Asia, generally endogenous endophthalmitis is usually caused by Gram negative organism *Klebsiella pneumonia* (KP), whereas gram positive organisms and fungi are the common agents seen in North America and Europe.[3] *Burkholderia cepacia* is a gram negative, oxidase positive, non- fermenting bacillus. Ocular infections in the form of Keratitis and endophthalmitis due to this organism is rarely reported. It was isolated in 1.8% cases of culture positive cases of endophthalmitis.[4] We report the first case of endogenous endophthalmitis following *B. cepacia* infection.

Case Report

A 33-year-old male, presented with the complaints of blurring of vision in the right eye of 3 days duration. Initial examination revealed visual acuity of 10/200 with no further improvement in the right eye and 20/20 unaided in the left eye. Anterior segment examination showed fresh keratic precipitates, grade 1 cells and flare in the anterior chamber, cells and exudates in the vitreous in the right eye. Examination of the retina revealed multiple intraretinal haemorrhages and

Cite this article as: Kurumkattil R, Trehan HS, Tandel K, Sharma VK, Dhar SK, Mahapatra T. Endogenous endophthalmitis secondary to *Burkholderia cepacia*: A rare presentation. Indian J Ophthalmol 2020;68:2283-5.

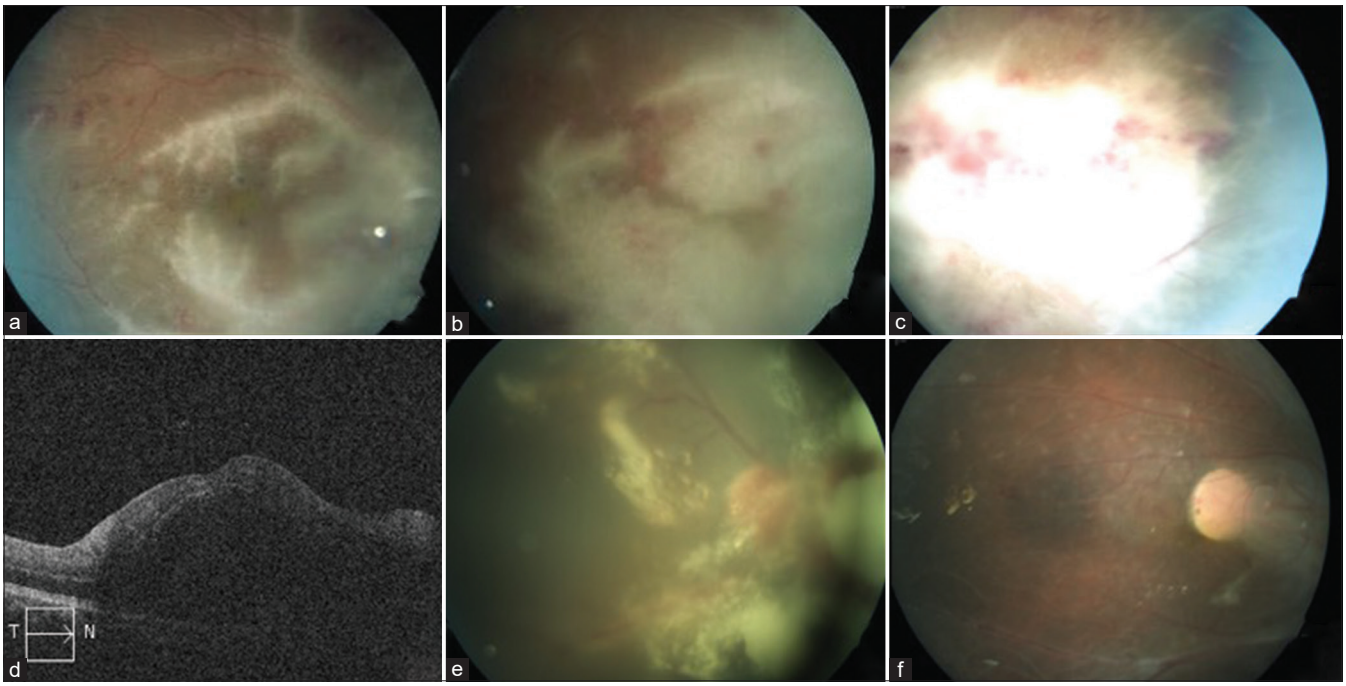

Figure 1: (a and b) Vasculitis and retinitis along superior and inferior arcade in the right eye, (c) Progression of the retinal necrosis and retinal abscess nasally in the right eye (d) Optical coherence tomography of Right eye showing increased retinal thickness, intra-retinal nerve fibre layer edema (e) Post - operative image showing Silicone filled eye and retinal traction nasally (f) fundus image after 12 months

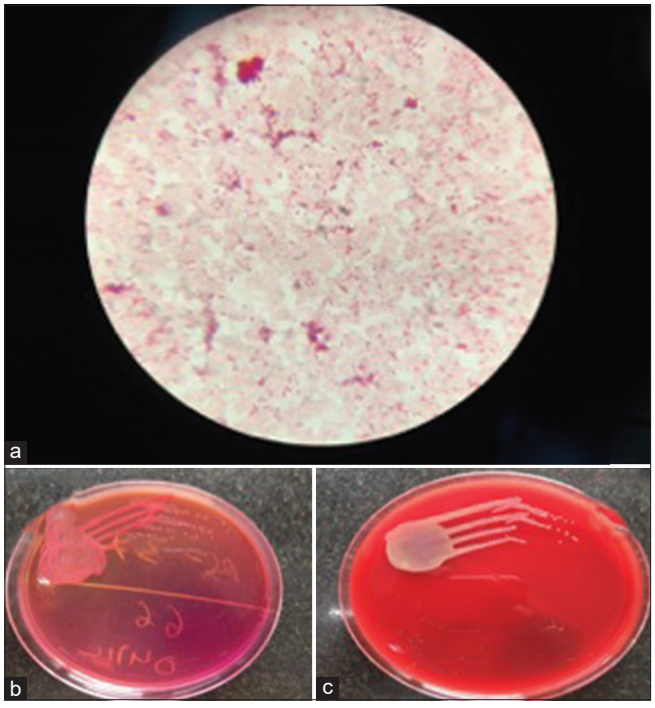

Figure 2: (a) Gram negative bacilli on Gram stain (b) Blood agar (c) Mac-Conkey agar showing bacterial Colony

retinitis along the supero-temporal, infero-temporal [Fig. 1a and b] and infero-nasal arcade and hyperemia of the optic disc and intraretinal edema of the same eye [Fig. 1c and d]. He gave history of taking intravenous fluids and medication from elsewhere for fever and loose motions 10 days before onset of his ocular symptoms. After admitted in the hospital he was

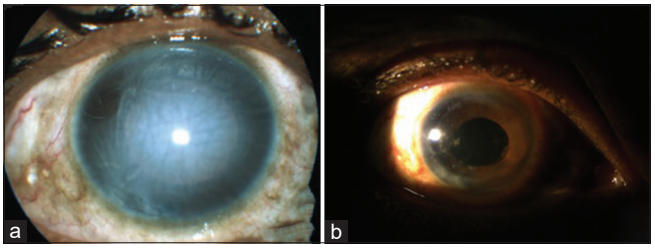

Figure 3: (a) corneal decompensation after repeated surgeries and Silicone oil (b) post DSAEK image showing clear cornea

investigated for the same. All investigations including HIV, TORCH, VDRL and blood culture had come negative. Vitreous biopsy was done and intravitreal clindamycin, and ganciclovir injection was given and which was repeated after 48 hours, keeping Toxoplasmosis and CMV retinitis as differential diagnosis along with topical steroids and cycloplegic eye drops. Intravenous Injection (IV) Acyclovir 500 mg TDS and Tab Septran DS BD was also started systemically. Over the next 3 days, he had further deterioration of vision in the right eye and it had gone down to perception of light with accurate projection of rays. Since there was not much improvement in his clinical condition, patient was taken up for vitrectomy. Vitreous sample was sent for gram stain, culture and antibiotic sensitivity test (AST). Intraoperatively 2 Disc Diameter size intra retinal abscess was noticed nasal to the optic disc [Fig. 1c], drainage of which was done and microbiological analysis, came positive for Gram-bacilli, and culture revealed *Burkholderia cepacia* on blood agar and Mac Conkey agar [Fig. 2a-c]. The sensitivity done on VITEK 2c system, confirmed the bacterial species which was only sensitive to the drug Meropenem, Levofloxacin and Amikacin. Intravenous Meropenum was started at a dose of 1 gm, 8

hourly along with Tab Levofloxacin 750 mg which was given for 7 days. Meropenem (50 µgm/0.1 ml) was injected inside the abscess through pars plana route and intravitreal injection Meropenem (50 µgm/0.1 ml) and Amikacin (400 µgm/0.1 ml) was repeated every 72 hrs, thrice. Though there was no change in his visual acuity and size of the retinal abscess, the exudates on the retina reduced. Persisting traction along superior and inferior arcade and retinal detachment was seen inferiorly, for which he was again operated. Initially, he was left under Perfluoro-N octane. After one week, this was exchanged with Silicone oil [Fig. 1e]. Patient had developed corneal decompensation due to repeated surgeries and Silicone oil inside the eye, for which a Descemet's Stripping automated endothelial keratoplasty (DSAEK) was done [Fig. 3a and b]. Patient's clinical condition has improved. During follow-up over 12 months, his retina is remaining attached with clear corneal graft [Fig. 1f]. There was no recurrence of infection.

Discussion

Endogenous endophthalmitis is rare and accounts for approximately 2-15% of all cases of endophthalmitis.[1] Endogenous endophthalmitis occurs when infectious organisms are spread from distant focus to the eye through the blood stream.[2] in Asia, generally endogenous endophthalmitis is usually caused by Gram negative organism *Klebsiella pneumonia* (KP), whereas gram positive organisms and fungi are the common agents seen in North America and Europe.[3] *Burkholderia cepacia* is a gram negative, oxidase positive, non-fermenting bacillus. Ocular infections in the form of Keratitis and endophthalmitis due to this organism is rarely reported. This organism is of low virulence, found in various aquatic environments[5,6] and found to be a colonizer in the irrigating solutions, nebulizer, intravenous fluids and topical anesthetic eye drops used in the hospitals.[7-9] It rarely infects a healthy individual. It is known to cause pneumonia, skin and soft tissue infection and genitourinary tract infection in patients of cystic fibrosis, individuals on hemodialysis, and chemotherapy.[7] If the patients are infected by a spread through the blood stream, it may result in gram-negative bacteremia.

Ocular manifestation of infection caused by *B. cepacia* is not well understood. Usually infection due to this organism is reported after cataract surgery, penetrating keratoplasty and post traumatic cases. Endophthalmitis due to *B. cepacia* is very rare. It was isolated in 1.8% cases of culture positive cases of endophthalmitis.[4]

We report the first case of endogenous endophthalmitis following *B. cepacia* infection Treatment of such cases remains a big challenge as these organisms are resistant to routine antimicrobials used to treat endophthalmitis and poor ocular penetration of drug inside the eye. Various studies have reported that susceptibility to antibiotics like ceftazidime, ciprofloxacin and amikacin. Seeing the clinical findings, patient was started on Injection acyclovir and Tab Septran, and later on intravenous and intralesional Meropenum based on the antibiotic sensitivity reports, and patient responded to the treatment.

Conclusion

Infection due to *Burkholderia cepacia* can preset as acute or delayed post- operative or posttraumatic endophthalmitis. Endogenous endophthalmitis due to Burkholderia cepacia, that too where it is presented as retinal abscess and retinal vasculitis is extremely rare and never reported before. Our aim was to highlight such a case treated at our center.

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.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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