

# ***Burkholderia Cepacia* Orbital Cellulitis Causing Blindness: A Case Report.**

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## **ABSTRACT**

*Burkholderia cepacia* is an aerobic gram-negative bacillus with multidrug resistance found in various moist environments. It has been reported in keratitis and endophthalmitis. We described a case of *Burkholderia cepacia* orbital cellulitis, which believed to be the first reported case. A 71-year-old gentleman with underlying poorly controlled Type 2 diabetes mellitus and hypertension, presented with left eye sudden blindness and complete ptosis for one day. On examination, left eye was non-perceptive to light with positive relative afferent pupillary defect, mild proptosis, chemosis, complete ptosis and total ophthalmoplegia. Eye swab culture revealed *Burkholderia cepacia*. Culture for fungal growth was negative. Despite treatment with intravenous cloxacillin, ceftriaxone and metronidazole, and later vancomycin, the nerve dysfunction and visual loss were irreversible. Bacterial orbital cellulitis rarely presents with blindness. This case represents the virulent nature of *Burkholderia cepacia*.

**Keywords:** *Burkholderia cepacia*, blindness, orbital cellulitis, ophthalmoplegia, virulence

## **INTRODUCTION**

Orbital cellulitis is an acute infection of the soft tissue around the eye. It is caused by bacteria or fungus with potentially serious complications including blindness, meningitis, cavernous sinus thrombosis, and intracranial abscess formation.<sup>1-3</sup> The most common causative bacteria are *Streptococcus* species, *Staphylococcus aureus*, and *Haemophilus in-*

*fluenza*, whereas *Pseudomonas*, *Klebsiella*, *Enterococcus* and *Eikenella* are less common organisms.<sup>3,4</sup> We described the first reported case of orbital cellulitis caused by *Burkholderia cepacia* and its virulent nature that leads to blindness.

## **CASE REPORT**

A 71-year-old gentleman with underlying poorly controlled Type 2 diabetes mellitus and hypertension with no previous eye complaints, presented with left eye blindness and complete ptosis. It was preceded with left eye progressive blurred vision and eyelid swelling for 2 days. He denied diplopia, ocular trauma nor insect bite on the eyelid and did not have

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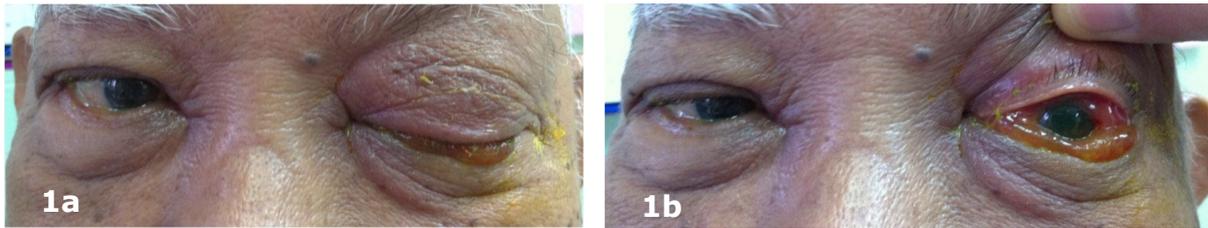


Figure 1. (a) Left eye complete ptosis with (b) mild proptosis and extensive chemosis.

any fever. On examination, there was no perception to light in the left eye with positive relative afferent pupillary defect. Other examination revealed mild proptosis with 2 mm difference, mild lid erythema and edema, chemosis, complete ptosis and total ophthalmoplegia [Figure 1(a) and (b)]. Ocular examination was unremarkable except cataract and mild non-proliferative diabetic retinopathy. Eye swab culture was taken and this grew *Burkholderia cepacia*. Blood culture for bacterial and fungal growth was negative. Computed tomography of orbit and paranasal sinuses did not show evidence of subperiosteal abscess or sinusitis [Figure 2(a) and (b)]. Endoscopic examination of nasal passages by otolaryngology team did not reveal sinusitis, or necrotic tissue that was suggestive of fungal infection. The patient received intravenous cloxacillin, ceftriaxone and metronidazole for 1 week. However in view of poor response, cloxacillin was changed to vancomycin on Day 3 of treatment for more empirical coverage. Subsequently the chemosis and proptosis re-

solved following antibiotic treatment. However, patient did not recover vision in his left eye, with complete ptosis and total ophthalmoplegia at 1 year of follow up.

## DISCUSSION

Orbital cellulitis is most commonly caused by adjacent sinusitis, accounting for more than 90% of all cases.<sup>1</sup> It can be caused by direct extension of infection from the globe, eyelids, or periocular tissues. Bacterial orbital cellulitis usually presents with marked proptosis, ophthalmoplegia and decreased vision.<sup>3</sup> It rarely presents with complete loss of vision as is our case. Severe visual impairment and rapid deterioration are mostly seen in orbital cellulitis caused by mucormycosis; however such clinical picture has been reported in bacterial infection caused by necrotizing strains of *Streptococcus pyogenes* and methicillin-resistant *Staphylococcus aureus*.<sup>5, 6</sup>

In our patient, eye swab culture was



Figure 2. Contrast computed tomography of orbit and paranasal sinuses revealed (a) minimal left eye proptosis (b) but did not show evidence of subperiosteal abscess or sinusitis.

the only positive culture which grew *Burkholderia cepacia*. *Burkholderia cepacia* (previously known as *Pseudomonas cepacia*) is a gram-negative, oxidase-positive, non-fermentative bacillus.<sup>7</sup> It can be found in soil, water, and infected plants, animals, and humans.<sup>8</sup> It has significant agricultural uses for promoting crop growth. In Malaysia, *Burkholderia cepacia* can be found in oil palm roots.<sup>9</sup> Our patient lives in a residential area near an oil palm estate and it is possible that he may have acquired the infection from there.

Patients may acquire *Burkholderia cepacia* from the environment or through patient-to-patient transmission.<sup>10</sup> It has become a significant opportunistic pathogen in immunosuppressed patients, causing the fatal "Cepacia Syndrome", especially in patients with cystic fibrosis. However, it is seldom found outside the lungs or in immunocompetent hosts. Ocular infections caused by *Burkholderia cepacia* are rare. To date, cases due to this gram-negative organism were reported in keratitis and endophthalmitis.<sup>7, 11-13</sup> Sachdeva *et al* reported that *Burkholderia cepacia* endophthalmitis accounts for 1.8% of culture positive endophthalmitis cases.<sup>12</sup> It can present as post-traumatic, acute-onset and delayed-onset post-operative endophthalmitis; and is associated with poor visual outcomes.<sup>12</sup>

*Burkholderia cepacia* is a highly virulent multidrug resistant organism.<sup>14</sup> It possesses antibiotic efflux pumps to remove antibiotic from the cell and forms biofilms to decrease contact to antibiotic. It also acquires beta-lactamases and altered penicillin-binding proteins. Because of multiple virulent factors, it has developed resistance to a number of antibiotics such as aminoglycosides, first- and second-generation cephalosporins, anti-pseudomonal penicillins, polymyxins, chloramphenicol, trimethoprim and fluoroquinolones.<sup>10</sup> Despite the multidrug resistance

towards common ophthalmic medication, cases of successful treatment were reported but rare.<sup>15, 16</sup> A patient with *Burkholderia cepacia* post-operative endophthalmitis with initial visual acuity of hand movement was able to achieve 6/6 vision after vitrectomy and five intravitreal antibiotic injection including ceftazidime and amikacin.<sup>15</sup> Another patient with *Burkholderia cepacia* keratitis following laser-assisted in situ keratomileusis (LASIK) was successfully treated with topical imipenem-cilastatin and polymyxin B/trimethoprim ophthalmic solution.<sup>16</sup>

In addition, *Burkholderia cepacia* can survive in aqueous environments such as a variety of solutions, medications, intravenous fluids, and even disinfectants and antiseptics such as benzalkonium chloride and chlorhexidine.<sup>17</sup> It is also unaffected by many preservatives including Betadine. Two studies reported that acute post-operative endophthalmitis following uneventful cataract surgery are due to *Burkholderia cepacia* from contaminated anaesthetic eye drops and contaminated trypan blue dye.<sup>13, 18</sup> In view of this, *Burkholderia cepacia* contamination test must be conducted on pharmaceutical products to ensure the absence of *Burkholderia cepacia*.

Fungal infection of the orbits such as Mucomycosis generally presents with more severe visual impairment and rapid progression.<sup>19</sup> Severe symptoms such as eyelid edema, proptosis, visual loss, pain and orbital apex syndrome involving cranial nerves II, III, IV, V1, and VI, and orbital sympathetics have been reported in Mucomycosis. It may lead to nasal and palatal necrosis, hence biopsy obtained from necrotic tissue is essential for timely diagnosis. Fungal orbital cellulitis has a high mortality rate in patients who are immunocompromised.<sup>20</sup> Thus, in patients with orbital cellulitis that associated with rapid clinical deterioration and visual loss, one should not neglect the possibility of fungal infection.

Work up including imaging and co-management with otorhinolaryngology team are essential for prompt diagnosis. In cases suspicious of fungal infection, a more aggressive approach to look for necrotic tissues at nasal passages and palate is crucial for the diagnosis of mucormycosis. In our case, the patient sustained blindness following virulent *Burkholderia cepacia* infection, which we postulated that he acquired infection from his residential environment. Combined with his poor sugar control, possibly led to an immunosuppressed state that promoted rapid and severe orbital infection. The mechanism of visual loss in the presented eye remained unclear. The severe infection may have caused ischemic necrosis, bacterial invasion, compression and stretching of the retrobulbar optic nerve.<sup>2</sup>

## CONCLUSION

In conclusion, bacterial orbital cellulitis that presents with rapidly decreased vision is rare. High level of suspicion for fungal causes must be maintained especially in immunosuppressed patients. In spite of poor visual outcome, aggressive treatment in orbital cellulitis is needed to prevent intracranial extension that is life threatening. This case demonstrated the virulent nature of *Burkholderia cepacia*.

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