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Ocular melioidosis: a diagnostic challenge with devastating visual consequences

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Abstract

Background: Melioidosis is a multi-system infectious disease caused by *Burkholderia pseudomallei*. Ocular involvement in melioidosis is rare, with a reported prevalence of less than 2%. It frequently affects adults with diabetes. For severe melioidosis, ceftazidime remains the first-line treatment, followed by extended maintenance therapy to reduce the risk of relapse and recurrence.
Case presentation: A 58-year-old man, presented with a 4-day history of pain, redness and photophobia in the right eye. He had diabetes with a history of disseminated melioidosis. Ocular examination showed conjunctival injection, hypopyon in the anterior chamber, and elevated intraocular pressure in the right eye. A conjunctival culture and sensitivity swab found *Burkholderia pseudomallei*. He was treated with intravenous ceftazidime followed by oral trimethoprim-sulfamethoxazole and responded well to treatment.
Conclusion: Early diagnosis guided by a high index of clinical suspicion as well as prompt treatment are crucial for favorable outcomes.

Keywords: Burkholderia pseudomallei, endophthalmitis, ocular melioidosis

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Melioidosis okular: cabaran diagnostik dengan kesan buruk terhadap penglihatan

Abstrak

Latar belakang: Meliodosis merupakan penyakit berjangkit multisistem yang disebabkan oleh Burkholderia pseudomallei. Penglibatan mata dalam meliodosis jarang berlaku, dengan prevalens yang dilaporkan kurang daripada 2%. Ia sering menyerang golongan dewasa yang mempunyai penyakit kencing manis. Untuk penyakit meliodosis yang serius, ceftazidime kekal sebagai rawatan utama, diikuti dengan terapi jangka masa panjang bagi mengurangkan risiko penyakit berulang. *Laporan kes:* Seorang lelaki berumur 58 tahun hadir dengan sejarah sakit pada mata kanan, kemerahan dan fotofobia apabila melihat cahaya selama empat hari. Beliau merupakan pesakit kencing manis dengan sejarah meliodosis diseminata. Pemeriksaan mata kanan menunjukkan konjunktiva merah, hipopion di ruang hadapan mata dan tekanan mata yang tinggi. Kultur dan ujian sensitiviti swab konjunktiva mata kanan mengenal pasti Burkholderia pseudomallei. Pesakit telah dirawat dengan ceftazidime secara intravena, diikuti dengan trimetoprim-sulfametoksazol oral dan menunjukkan tindak balas yang baik terhadap rawatan. Kesimpulan: Diagnosis awal yang dipandu oleh tahap kecurigaan klinikal yang tinggi serta rawatan yang segera adalah penting untuk mencapai hasil rawatan yang baik.

Kata kunci: Burkholderia pseudomallei, endoftalmitis, melioidosis okular

Introduction

Melioidosis is an infectious disease caused by the gram-negative, motile, non-spore forming *Burkholderia pseudomallei* bacillus, which is found in soil and surface water and is widely distributed in tropical and subtropical regions. Although ocular involvement in melioidosis is rare, it may lead to devastating outcomes. The prevalence was estimated around 0.49 to 1.02%.[1] It is endemic in Southeast Asia (notably Thailand and Malaysia) and northern Australia.[2,3] Here, we report a case report of patient with melioidosis and ocular involvement.

Case report

A 58-year-old agricultural worker presented with a 4-day history of pain, redness, and photophobia in the right eye. Prior to this episode, he had poor baseline visual acuity, limited to counting fingers in both eyes, secondary to advanced diabetic eye disease with bilateral vitreous haemorrhage. His past medical history was significant for diabetes mellitus and disseminated melioidosis with splenic and left wrist abscess in 2023, microbiologically confirmed from blood and wrist pus cultures growing *Burkholderia pseudomallei*. He completed 6 weeks of intravenous ceftazidime followed by 3 months of oral doxycycline.

On initial examination, he was systemically stable with normal vital signs. Visual acuity was hand movement in the right eye and counting fingers in the left eye. Ocular examination revealed conjunctival injection, corneal oedema, fibrin, blood clot, and a nasal hypopyon in the anterior chamber, with elevated intraocular pressure in the right eye. Fundus examination was not possible due to vitreous haemorrhage. B-scan ultrasonography confirmed vitreous haemorrhage. A steroid challenge was performed and showed no worsening; thus, he was initially managed as neovascular glaucoma with severe anterior uveitis.

He was admitted for intraocular pressure control and further evaluation. Initial blood investigations, including full blood count, renal profile, liver function tests, and Mantoux test, were within normal limits. Endogenous endophthalmitis secondary to melioidosis was suspected in view of his diabetes mellitus, occupational exposure, prior history of melioidosis, and the appearance of vitreous loculations on follow-up B-scan imaging. He subsequently underwent intravitreal tap and injection of vancomycin and ceftazidime. Both vitreous and blood cultures yielded no growth. Conjunctival swab culture, however, grew *Burkholderia pseudomallei*. Abdominal ultrasonography revealed multiple splenic lesions, with the largest measuring approximately 0.9 × 1.2 cm, compared to a previous lesion measuring 1.5 × 2.0 cm during his prior episode of melioidosis in 2023.

One week later, a scleral abscess developed in the nasal aspect of the right eye (Fig. 1). Repeat B-scan showed improving vitreous loculations; however, a superior subretinal abscess was noted. He was referred for vitreoretinal intervention to reduce the intraocular infective load and improve antimicrobial penetration, but the patient declined the procedure.

He completed 8 weeks of intravenous ceftazidime, followed by eradication therapy with trimethoprim-sulfamethoxazole for 1 year. During follow-up, he remained well with no signs of disease recurrence, but he remained legally blind.

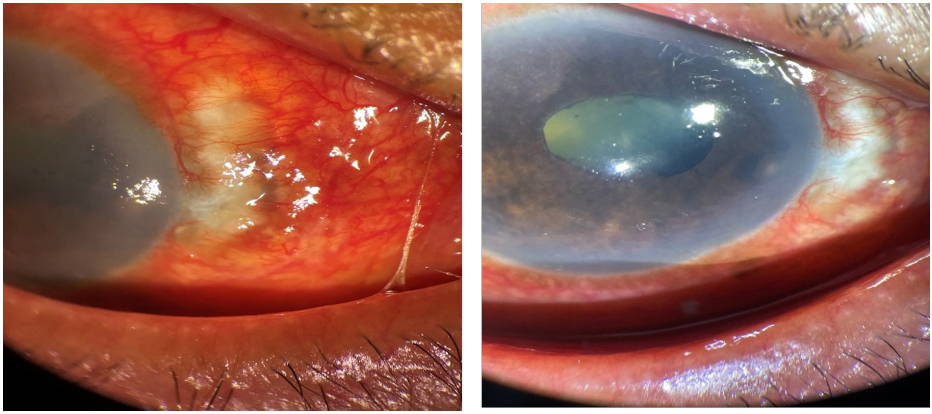

Fig. 1. Anterior segment photographs showing scleral abscess over nasal part of right eye.

Discussion

Melioidosis is a multiorgan infectious disease caused by *Burkholderia pseudomallei*. Based on previous reports, diabetes mellitus was the most frequently identified predisposing risk factor in Malaysia, with up to 60% of cases having pre-existing or newly diagnosed type 2 diabetes.[4] Recrudescence from latent infection may be triggered by impaired immunity, such as tuberculosis or acquired immunodeficiency syndrome.[5] These findings are consistent with our case report, which highlights disease recurrence in an older patient with diabetes and a prior history of disseminated melioidosis.

Melioidosis is widely recognized as “the great mimicker” because of its ability to affect any organ and present with a broad spectrum of clinical manifestations, ranging from benign skin and soft tissue lesions to a rapidly fulminant and fatal septicaemia. Ocular involvement can be either localized or as part of disseminated septicaemic melioidosis. Orbital cellulitis was the most common manifestation (44%) followed by endophthalmitis (25%), panophthalmitis (13%), preseptal cellulitis (13%), and panuveitis (6%).[6]

The gold standard for melioidosis diagnosis is isolation of *Burkholderia pseudomallei* from clinical specimens. Serological testing may aid presumptive diagnosis in severe septicaemic melioidosis cases or when infections are deep seated and tissue sampling is challenging.[7] This case emphasizes the diagnostic challenge posed by ocular melioidosis and the need for a high index suspicion in endemic regions, particularly among agricultural workers with relevant occupational exposure.

Treatment of fulminant melioidosis includes high-dose intravenous ceftazidime (100–120 mg/kg/day in divided doses), followed by an extended oral eradication phase to prevent relapse.[8,9] Despite initiation of systemic antibiotic, the occurrence

of endogenous endophthalmitis would still be unpreventable. Although antimicrobial therapy has shown to be effective, melioidosis is still highly associated with high mortality due to severe sepsis and its complications.

In most cases, the definitive management of ocular cases is surgery, including incision and drainage, debridement, pars plana vitrectomy, and/or enucleation. Despite appropriate systemic and intravitreal antimicrobial therapy, our patient declined surgical intervention and was left legally blind, underscoring the therapeutic challenges and poor visual outcomes associated with delayed or incomplete management.

Conclusion

In summary, ophthalmologists should maintain a high index of suspicion for ocular melioidosis in patients with ocular infections that fail to respond to conventional antibiotic therapy, particularly in endemic regions such as Malaysia. Although rare, ocular melioidosis can result in severe and irreversible visual impairment. Prompt recognition and timely initiation of appropriate systemic antimicrobial therapy, with or without surgical intervention, remain critical for optimizing visual outcomes and reducing mortality.

Declarations

Informed consent for publication
The patients provided informed consent for the publication of the clinical data and images contained in this case report.

Competing interests
None to declare.

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