Brunei International Medical Journal

OFFICIAL PUBLICATION OF
THE MINISTRY OF HEALTH
AND
UNIVERSITI BRUNEI DARUSSALAM

Volume 18 22 April 2022 (20 Ramadan 1443H)

DACRYOADENITIS WITH MULTIPLE UNILATERAL OCULAR ABSCESSES IN DISSEMINATED MELIOIDOSIS.

SYLVES P[1], SEAH RB[2], CAROLINE B[2], YAP JY[1], SHUAIBAH AG[1], HANIDA H[3].

[1]Department of Ophthalmology, Faculty of Medicine and Health Sciences Universiti Malaysia Sabah, Jalan UMS, 88400 Kota Kinabalu, Sabah, Malaysia.
[2]Department of Ophthalmology, Queen Elizabeth Hospital, 88586 Kota Kinabalu, Sabah, Ministry of Health Malaysia.
[3]Oculoplastic and Reconstructive Unit, Department of Ophthalmology, Queen Elizabeth Hospital, 88586 Kota Kinabalu, Sabah, Ministry of Health Malaysia.

ABSTRACT

Melioidosis is caused by the gram-negative bacterium *Burkholderia pseudomallei* and has a wide range of manifestations in many organ systems. Ocular melioidosis is a rare manifestation of *Burkholderia pseudomallei* that commonly presents with orbital cellulitis rather than dacryoadenitis and multiple unilateral ocular abscesses, which are uncommon. The risk of blindness in ocular melioidosis is high because this bacterium is resistant to many antibiotics, requires a longer period of treatment with antibiotics and the diagnosis is not always straightforward. Here we describe a different presentation of ocular melioidosis manifested with left preseptal abscess, orbital cellulitis with abscess and dacryoadenitis in a 50-year-old man who was treated successfully with surgery and antibiotics, along with a brief review of the literature.

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Brunei Int Med J. *2022;18:79-83*

ISSN 1560 5876 Print
ISSN 2079 3146 Online

Online version of the journal is available at www.bimjonline.com

Brunei International Medical Journal (BIMJ)
Official Publication of
The Ministry of Health
and
Universiti Brunei Darussalam

ISSN 1560-5876 Print
ISSN 2079-3146 Online

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Acknowledgements

Only persons who have made substantial contributions but who do not fulfill the authorship criteria should be acknowledged.

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ABSTRACT

Melioidosis is caused by the gram-negative bacterium *Burkholderia pseudomallei* and has a wide range of manifestations in many organ systems. Ocular melioidosis is a rare manifestation of *Burkholderia pseudomallei* that commonly presents with orbital cellulitis rather than dacryoadenitis and multiple unilateral ocular abscesses, which are uncommon. The risk of blindness in ocular melioidosis is high because this bacterium is resistant to many antibiotics, requires a longer period of treatment with antibiotics and the diagnosis is not always straightforward. Here we describe a different presentation of ocular melioidosis manifested with left preseptal abscess, orbital cellulitis with abscess and dacryoadenitis in a 50-year-old man who was treated successfully with surgery and antibiotics, along with a brief review of the literature.

Keywords: Abscess, Burkholderia pseudomallei, Dacryoadenitis, Melioidosis, Ocular, Unilateral.

INTRODUCTION

Melioidosis is caused by a gram-negative bacterium called *Burkholderia pseudomallei*, which is endemic in Southeast Asia and Northern Australia.[1,2] It has a wide range of manifestations involving many organs, such as the lungs, genitourinary system, skin, liver, pancreas, spleen, brain and joints, as well as bone and ocular structures.[2-4] Ocular manifestation is rare, with prevalence estimated as 0.49–1.02%.[5] Even though ocular melioidosis is rare, the risk of blindness due to this infection is high and can lead to life-threatening conditions if not treated early because the bacterium is resistant to many antibiotics. Here we describe a different presentation of a case of disseminated melioidosis with ocular melioidosis manifested with left preseptal abscess, orbital cellulitis with abscess and dacryoadenitis in a patient who was treated successfully with surgery and antibiotics, along with a brief review of the literature.

Corresponding author: Dr. Sylves Patrick, Senior Lecturer and ophthalmologist, Department of Ophthalmology, Faculty of Medicine and Health Sciences, Universiti Malaysia Sabah (UMS), 88400, Kota Kinabalu, Sabah, Malaysia Tel: +6010-5093168; E-mail: **sylves@ums.edu.my**

CASE REPORT

A 50-year-old male farmer with underlying uncontrolled diabetes mellitus, hypertension, dyslipidaemia, end-stage renal failure, a history of old ischemic stroke and both eyes treated for proliferative diabetic retinopathy presented with three weeks of left upper eyelid swelling. The swelling was associated with left eye pain, redness and reduced vision. He also had lethargy and fever, with left knee pain and swelling. He had no double vision, upper respiratory symptoms, ear discharge, toothache or symptoms of increased intracranial pressure. He also had no history of trauma or insect bites and had not taken any immunosuppressive medication.

On examination, his Glasgow Coma Scale (GCS) was 15/15. On examination of the left eye, visual acuity (VA) was 6/24 (unaided) and intraocular pressure (IOP) was 55 mmHg. There was generalised erythematous upper eyelid swelling that was non-fluctuant and tender. The eyelid was fully ptotic (Figure 1a). There was hypoglobus with limited extraocular muscle movement in all gazes. The conjunctiva was inflamed and chemosed. However, there were no signs of optic nerve dysfunction. Anterior segment examination of the right eye was unremarkable. Both eyes' posterior segments showed multiple old laser marks of panretinal photocoagulation. Other systemic examinations were unremarkable except for the left scalp and knee joint redness and swelling.

Blood investigations showed sign of infection with elevated liver enzymes (transaminases). Viral hepatitis, retroviral and syphilis screenings were negative. Blood culture grew *Burkholderia pseudomallei.* Contrast-enhanced computed tomography (CECT) of the brain, orbit and paranasal sinuses showed left preseptal abscess, left orbital abscess, left frontal lobe abscess, left scalp abscess, left infratemporal abscess and left lacrimal gland enlargement with small well-defined hypodensities seen at bilateral internal capsules and left external capsule (Figure 2). Ultrasonography of the abdomen revealed no evidence of intra-abdominal abscess. Left knee ultrasonography showed heterogeneous hypoechoic synovial fluid.

Initially, the patient was treated with intravenous metronidazole and augmentin but the condition did not improve. Blood culture grew Burkholderia pseudomallei after 48 hrs and he was diagnosed with disseminated melioidosis with transaminitis. His antibiotics were immediately changed to intravenous ceftazidime (2 g od) and meropenem (2 g bd). He was co-managed by multi-disciplinary teams consisting of internal medicine, neurosurgical, otorhinolaryngology (ORL), orthopaedic and ophthalmology teams. Bedside aspiration of the scalp abscess was performed by the neurosurgical team but the left frontal lobe and left infratemporal abscesses were treated conservatively by the neurosurgical and ORL teams, respectively. The left knee underwent an arthrotomy washout performed by the orthopaedic team. He underwent incision and drainage of the preseptal and orbital abscesses via sub-brow anterior orbitotomy (Figure 1b) performed by the ophalthalmolo-

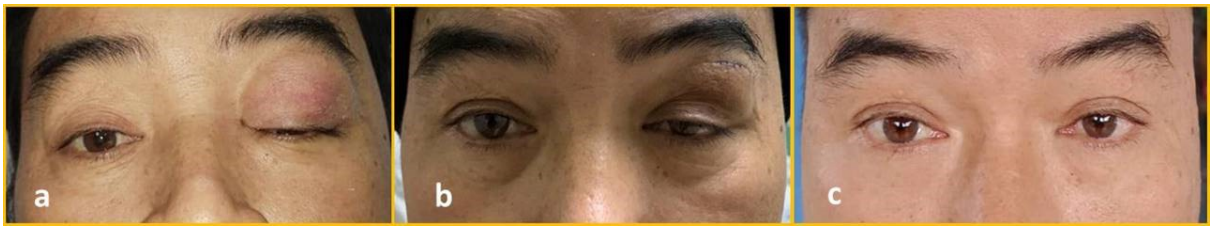

Figure 1: Dacroadenitis and ocular swelling at a: Initial assessment; b: Postoperative Day 3; c: After 6 months of follow-up. (Click to enlarge)

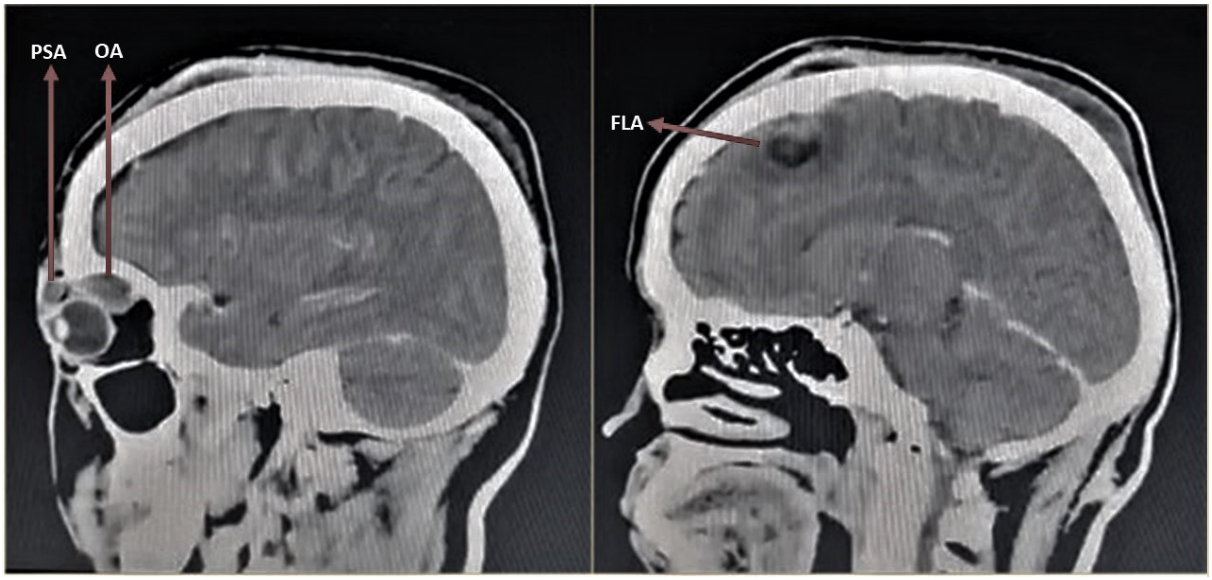

Figure 2: Contrast-enhanced computed tomography showing left preseptal (PSA), orbital (OA) and frontal lobe (FLA) abscesses. The orbital abscess displaced the left eyeball downwards.

gy team. Pus from all the sites above (scalp, knee and orbital) grew *Burkholderia pseudomallei.* He was also treated with moxifloxacin eyedrops every 4 h and with IOP-lowering eyedrops.

For diabetic controlled throughout the admission, he was treated with subcutaneous insulin (Mixtard, 14/10 units bd) and his other prescribed medications included amlodipine, 10 mg od; metoprolol, 25 mg bd; aspirin, 150 mg od; pantoprazole, 40 mg od; and hematinics. The intravenous ceftazidime was continued for 8 weeks and the meropenem for 3 weeks, along with trimethoprim/ sulfamethoxazole (80 mg/400 mg, three tablets a day) for 6 months.

After completion of treatment, the patient's liver function test normalised. Repeated CECT of the brain and orbit/paranasal sinuses showed resolved left preseptal, orbital, frontal lobe, scalp and infratemporal abscesses, with normal left lacrimal gland. His left eye swelling had resolved completely with no more ophthalmoplegia and with a best-corrected VA of 6/6 (Figure 1c).

DISCUSSION

The risk factors for ocular melioidosis are being male, a farmer, having diabetes mellitus and having renal disease.[5] All these risk factors were present in our case, in addition to the fact that the case occurred in an endemic region in Sabah, Malaysia. *Burkholderia pseudomallei* can be found in soil and surface water.[6] The patient may have acquired the infection via direct contact with contaminated soil or surface waters, especially through skin abrasions that occur during farming.[3]

Ocular melioidosis can be localised or part of disseminated melioidosis as was the case represented here. Disseminated melioidosis with ocular melioidosis has been reported in about 56% of cases.[5] Ocular melioidosis can be manifested as preseptal cellulitis, eyelid abscess, orbital cellulitis, orbital abscess, corneal ulcer, endophthalmitis, panophthalmitis, dacryocystitis, panuveitis, subretinal abscess and subconjunctival abscess.[5,7] However, the most common manifestation is orbital cellulitis, followed by preseptal cellulitis, endophthalmitis, panophthalmitis and panuveitis.[5]

Unilateral multiple ocular abscesses

are uncommon in ocular melioidosis. A case of bilateral multiple ocular abscesses was reported, manifested with bilateral orbital and eyelid abscesses that were successfully treated with surgical drainage and antibiotics. However, this was in a child with presumed ocular melioidosis.[8] Yaisawang et al., reported a case of culture-confirmed disseminated ocular melioidosis manifested with unilateral multiple orbital abscesses and necrotising fasciitis, with good outcome.[5] However, surgical drainage of the abscesses was not needed and the final visual outcome was not recorded.[5]

Unilateral multiple subconjunctival abscesses can occur in ocular melioidosis but such manifestation is also likely in other bacterial infections.[7] In our case, the abscesses was present in two different compartments (preseptal and orbital) of the eye were significant enough to need surgical drainage, which is unlikely to be the case in other bacterial infections. Knowledge of this manifestation will help ophthalmologists to prepare for melioidosis in those cases where there is no response to the standard antibiotic treatment.

Lacrimal gland involvement in melioidosis has been reported in the paediatric population in Sarawak, Malaysia.[9] However, this article needs to be interpreted cautiously because the authors described the lacrimal gland involvement as an inflamed tender swelling at the medial aspect of the lower eyelid, which refers more to dacryocystitis than dacryoadenitis. In our case, the left lacrimal gland was enlarged, as seen from the CECT, and this normalised after completion of treatment, suggesting lacrimal gland involvement in disseminated melioidosis.

A retrospective study conducted by Yaisawang et al. reported that about 73% of ocular melioidosis patients presented with significant visual impairment or blindness.[5] Even with adequate surgical intervention, 64% and 14% ended up legally blind and with enucleation, respectively.[5] This indicates that aggressive intervention is crucial in preserving the vision and saving the eye without undue delay. In our case, the plan for early surgical intervention was very important in relieving the high IOP, which can lead to blindness from irreversible glaucomatous optic neuropathy. After treatment with IOP-lowering agents, prolonged systemic antibiotics and surgical intervention, the visual and cosmetic outcomes were good.

CONCLUSION

In conclusion, ocular melioidosis with multiple unilateral abscesses and dacryoadenitis is uncommon and can present as part of disseminated melioidosis, which carries a high risk of blindness and can even lead to mortality. The diagnosis is not always straightforward, which delays treatment and further worsens the prognosis. However, it is curable, with good outcomes following the appropriate surgical intervention and antibiotic treatment. Ophthalmologists need to be aware of the different ocular manifestations so that the appropriate treatment can be commenced without undue delay.

CONFLICT OF INTEREST

The author reported no conflict of interest or financial liability.

INFORMED CONSENT

Informed consent has been obtained from the patient in regards to the pictures and details included in this report.

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