



Case report

Periorbital cellulitis and eyelid abscess as ocular manifestations of melioidosis: A report of three cases in Sarawak, Malaysian Borneo



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ABSTRACT

Melioidosis is an infection caused by the Gram-negative bacterium *Burkholderia pseudomallei*. Melioidosis can affect any organ and result in a broad spectrum of diseases. Ocular involvement in melioidosis is rare but can potentially lead to devastating outcomes. We describe ocular manifestations in 3 patients diagnosed with culture-confirmed melioidosis in Kapit, Sarawak, Malaysian Borneo. Melioidosis affecting the eye can present as periorbital cellulitis and eyelid abscess. High clinical suspicion and early empirical treatment were crucial to a successful outcome. Surgical drainage of the abscess was an important part of management beyond standard antimicrobial therapy.

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Introduction

Melioidosis is caused by *B. pseudomallei*, which is a facultative intracellular Gram-negative, saprophytic bacterium, commonly found in soil or contaminated water [1]. It is an endemic disease in northern Australia and Southeast Asian countries, especially Thailand and Malaysia. Melioidosis can virtually affect any organ in the body and present with diverse clinical manifestations, including pneumonia, genitourinary infection, skin and soft tissue infection, internal organ abscesses, septic arthritis, neurological melioidosis, and fulminant septicaemia without evident focus [2]. Despite improvements in antimicrobial therapy, melioidosis is still associated with high mortality attributable to severe sepsis and its complications [3].

Ocular involvement in melioidosis is rare and the prevalence was estimated at only 0.49–1.02 % [4]. A retrospective review of 16 cases of ocular involvement in melioidosis in Thailand revealed orbital cellulitis as the most common manifestation (7 cases) followed by endophthalmitis (4 cases), preseptal cellulitis (2 cases), panophthalmitis (2 cases), and panuveitis (1 case) [4]. Here, we report a case series of 3 patients with melioidosis and ocular involvement in Kapit, Sarawak, Malaysian Borneo.

Case history

Case 1

A 23-year-old housewife, previously well, presented with a one-week history of fever and right lower eyelid swelling. She denied any blurring of vision, eye pain or eye discharge. Upon arrival at the hospital, she was comfortable with normal vital signs. Physical examination revealed an abscess at right lower eyelid, measuring 1.5 × 1 cm (Fig. 1), followed by a detailed ophthalmic examination that did not reveal any abnormality. A bedside needle aspiration of the abscess was performed in which 2 mL of purulent material was aspirated and sent for culture. She was noted to have mild anemia (hemoglobin, 10.3 g/dL), thrombocytopenia (platelet, 46×10^3 /uL), and raised serum transaminases (AST 48 U/L, ALT 73 U/L). The chest radiograph showed mild bilateral lung infiltrates and abdominal ultrasonography revealed multiple splenic micro-abscesses. The patient was administered with intravenous ceftazidime 2 g 8-hly as an empirical treatment for melioidosis, in view of pneumonia and formation of abscesses in the spleen and eyelid.

On the third day of hospitalization, she developed worsening respiratory distress that required endotracheal intubation and mechanical ventilation. A chest radiograph was repeated and showed worsening lung consolidation. *B. pseudomallei* was isolated from the patient's blood, sputum and purulent material from eyelid abscess. Her condition subsequently improved with antimicrobial therapy. She was extubated after a week and

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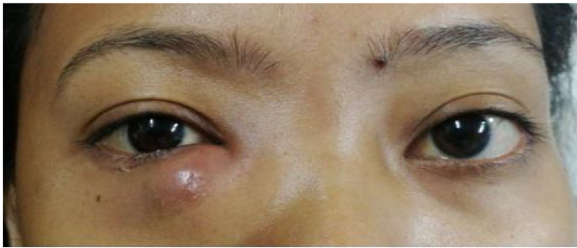


Fig. 1. Right lower eyelid abscess.

attained normalization of blood parameters. She completed intravenous ceftazidime for 4 weeks, followed by a 20-week course of eradication therapy consisting of trimethoprim-sulfamethoxazole and doxycycline. During a follow-up review in the medical clinic, she was well and the eyelid abscess resolved. A repeat abdominal ultrasonography post-treatment showed resolution of the splenic microabscesses.

Case 2

A 62-year-old male farmer presented with a two-week history of fever, associated with cough, dyspnoea and progressive right eye swelling of one-week duration. His medical history included diabetes mellitus and ischaemic heart disease. Vital signs were normal upon arrival at the hospital and his physical examination revealed right periorbital swelling (Fig. 2) while ophthalmic examination was normal except mild diabetic retinopathy changes. The initial working diagnosis was right periorbital cellulitis. The full blood count was normal while biochemistry panel revealed normal renal function and elevated liver enzymes (AST 78 U/L, ALT 64 U/L). A chest radiograph showed bilateral perihilar infiltrates while abdominal ultrasonography revealed multiple splenic microabscesses. Melioidosis was suspected due to presence of the risk factors of farming activity, diabetes mellitus and the finding of splenic microabscesses. Hence, intravenous ceftazidime was given as empirical treatment for melioidosis.

Despite early commencement of antimicrobial, the patient developed respiratory failure on the third day which required mechanical ventilatory support. Continuous infusion of noradrenaline was also started. He was intubated for 4 days in the intensive care unit and during this period, the antimicrobial regime was escalated to meropenem. Later, an abscess formed at the right upper eyelid for which incision and drainage was performed. Cultures of the blood and purulent material from right eyelid abscess yielded *B. pseudomallei*. Intravenous meropenem was changed back to ceftazidime as the bacterial isolate was sensitive to ceftazidime, and the patient has improved clinically. He continued to receive intravenous ceftazidime 2 g 8-hly for 4



Fig. 2. Right periorbital swelling involving the upper and lower eyelids.

weeks, followed by oral doxycycline and trimethoprim-sulfamethoxazole as eradication therapy. However, the latter was substituted with amoxicillin-clavulanate as he developed leukopenia. He recovered well, and the right eyelid and splenic abscesses resolved.

Case 3

A 58-year-old male farmer presented with fever and right eye swelling of one-week duration. His past medical history include diabetes mellitus and bacteraemic melioidosis in 2016 for which he was treated with intravenous ceftazidime and oral trimethoprim-sulfamethoxazole. Upon presentation at the hospital, his general condition was stable and the vital signs were normal. There was periorbital swelling of the right eye noted. The ophthalmic examination showed mild non-proliferative diabetic retinopathy in both eyes. Initial blood investigations including full blood count, renal and liver function tests were within the normal limits. The chest radiograph and abdominal ultrasonography did not reveal any abnormality. The initial impression made was right periorbital cellulitis and he was started on intravenous ampicillin-sulbactam 1.5 g 8-hly. However, persistent temperature spikes was observed over the next 3 days and suspicion for melioidosis was raised in view of diabetes mellitus, occupational exposure and previous history of melioidosis. The regime was escalated to intravenous ceftazidime 2 g 8-hly, to treat for melioidosis. Computed tomography of the orbit revealed right periorbital cellulitis (Fig. 3).

A week later, an abscess developed over the right upper eyelid. Incision and drainage of the abscess was performed where copious amount of purulent material was drained and sent for microbiological analysis and it and blood cultures taken on admission yielded *B. pseudomallei*. He completed intravenous ceftazidime for 2 weeks and responded to treatment whereby fever and right eye swelling subsequently resolved. He then completed eradication therapy consisting of trimethoprim-sulfamethoxazole and doxycycline for 20 weeks. During a follow-up review at the medical clinic, he was well and did not show any signs of disease recurrence.

Discussion

Melioidosis is a multiorgan infectious disease which can present with a wide spectrum of clinical presentations. Based on the Darwin study, pneumonia was found to be the most common presentation of melioidosis in approximately half of the cases. Less common presentations include genitourinary infection (14%), skin infection (13%), bacteraemia without evident focus (11%), septic arthritis or osteomyelitis (4%) and neurological melioidosis (3%) [5]. Ocular involvement in melioidosis is rare and the prevalence was estimated as only 0.49–1.02% [4]. There have been several case reports previously on corneal ulcers, orbital cellulitis and endophthalmitis caused by *B. pseudomallei* [6–8].

Yaisawang et al. in her series of 18 cases, reported orbital cellulitis as the most common manifestation of ocular melioidosis, followed by endophthalmitis, preseptal cellulitis, panophthalmitis, and panuveitis [4]. This is in contrary to our observation although only with 3 patients whereby eyelid abscess, either occurs in isolation or association with periorbital cellulitis was the most common presentation. However, there is one aspect in common as most cases of ocular melioidosis are associated with *B. pseudomallei* bacteraemia. This finding may be explainable by the nature of *B. pseudomallei* infection, which tends to present with bloodstream infection. We also observed that patients with ocular melioidosis are more likely to have disseminated septicemia with multiorgan involvement such as splenic abscesses and pneumonia.

Periorbital cellulitis, also known as preseptal cellulitis is an infection of the eyelid and superficial periorbital soft tissues



Fig. 3. Axial CT scan of orbit shows right periorbital swelling with soft tissue thickening (white arrow).

without the involvement of the globe and orbit. It typically presents with eyelid edema and erythema. There are three main routes for pathogen inoculation namely, direct inoculation, spread from contiguous structures e.g. paranasal sinuses and haematogenous [9]. *Staphylococcus aureus*, *Streptococcus pneumoniae*, other streptococci, and anaerobes are common causative organisms of periorbital cellulitis [10]. Periorbital cellulitis due to *B. pseudomallei* is rare and has only been described in a few case reports. Eyelid abscess is a common association with periorbital cellulitis, which was seen in two of our patients.

Patients with periorbital cellulitis generally do not require surgical intervention except in cases of an eyelid abscess. If present, needle aspiration or incision and drainage is usually performed and microbiologic evaluation should be performed, with culture and sensitivity results should be used to guide appropriate antimicrobial therapy [9]. In all 3 patients, cultures of blood and purulent material from the eyelid abscess yielded *B. pseudomallei*, grown on modified Francis culture medium without gentamicin, as the *B. pseudomallei* isolates from Sarawak are predominantly susceptible to aminoglycosides [11]. This was obtained after preliminary identification using API20NE test (Biomerieux) and Gram staining. Upon sensitivity testing using the E-test method, the organism was found to be susceptible to ceftazidime, amoxicillin-clavulanic acid, gentamicin, and trimethoprim-sulfamethoxazole. A confirmatory test by real-time PCR was however not available at our facility.

The majority of cases of periorbital cellulitis and eyelid abscess resolve without complication following antimicrobial therapy. Nevertheless, accurate diagnosis and expeditious treatment is crucial as the infection can extend posteriorly into the orbit causing orbital cellulitis, subperiosteal abscess or orbital abscess.

These will potentially lead to significant visual and central nervous system complications [9]. All 3 patients received an antimicrobial regime as per Darwin melioidosis guidelines [12]. The first and second patients received intensive phase treatment of 4 weeks duration in view of deep-seated splenic abscesses, while the third patient was treated with intensive phase treatment of 2 weeks duration for bacteraemia without deep-seated abscess. It should be highlighted that our patients received early administration of empirical antimicrobial therapy and responded well to treatment without any ophthalmic complications.

In conclusion, clinicians working in endemic areas should maintain a high index of clinical suspicion of melioidosis in patients presenting with eyelid abscess and periorbital cellulitis. Early empirical antimicrobial treatment should be commenced promptly in suspicious cases. Surgical drainage of abscess is an important part of management beside standard antimicrobial therapy.

Author Statement

CYC managed the case and wrote the paper.

Ethics approval and consent to participate

Written informed consent for publication of the clinical details was obtained from the patient.

Declaration of Competing Interest

The authors declare that they have no competing interests.

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