

RECURRENT SUPPURATIVE DACRYOCYSTITIS SECONDARY TO MELIODOSIS

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ABSTRACT

Background: We are here to report a rare case of acute suppurative dacryocystitis secondary to *Burkholderia pseudomallei*.

Materials and Methods: A case report

Result: A 70-year-old Malay women presented with recurrent episode of suppurative dacryocystitis that were resistant to oral and topical antibiotics. Culture and sensitivity swab from the fistula track yield *Burkholderia pseudomallei*. Patient was started on intravenous ceftazidime and topical ceftazidime 5% for two weeks and a course of oral trimethoprim/sulfamethoxazole for 6 weeks.

Conclusion: *Burkholderia pseudomallei* should be consider as a rare cause for acute suppurative dacryocystitis in South East Asia region. Systemic investigation and imaging is important to look for deep seated abscess in various organ.

Keywords: Meliodosis, dacryocystitis, *Burkholderia pseudomallei*

1.0 Introduction

Inflammation of the lacrimal sac and obstruction of the nasolacrimal duct can lead to the development of acute dacryocystitis.¹⁻² Stagnation of the fluid in the obstructed sac along with proliferation of bacteria is the most common pathophysiology of the formation of acute suppurative dacryocystitis.¹ Among the organism isolated in acute dacryocystitis, the most common organism found is gram positive organism, out of which, *Staphylococcal aureus* is the predominant microorganism isolated.¹⁻³ Although *Burkholderia pseudomallei* is known aetiology for skin and subcutaneous tissue abscess formation, so far it was never been reported as a microorganism related to the formation of acute dacryocystitis.

Therefore, we would like to report this rare case where *Burkholderia pseudomallei* was isolated as the causative organism for acute suppurative dacryocystitis in our patient.

2.0 Materials and Methods

A case reports

3.0 Result

A 70-year-old Malay lady presented with pain, redness and swelling of the right medial canthal for 4 days. It was associated with epiphora. She was diagnosed to have acute dacryocystitis and was treated with oral antibiotic for one-week. However, 2 weeks later, she presented again with recurrent episode of similar problem. This time it was associated with yellowish purulent discharge. There was no history of trauma or injury. There was no history of fever, upper respiratory tract infection, facial swelling or diplopia. Her vision was good in both eyes. She was diagnosed to have diabetic mellitus for more than 10 years and on oral hypoglycaemic agent. Her blood sugar control was good. She is a full time housewife and her hobby is gardening and flower planting.

On examination, there was presence of a poorly defined indurated area over the right medial canthal area, extending to middle half of the lower lid. The skin above it was erythematous, wrinkled and covered with crusted discharge. There was a fistula with purulent discharged upon compression. Both conjunctival and cornea were normal. She was diagnosed to have recurrent dacryocystitis with fistula.

Patient was treated with oral cloxacillin 500 mg four times a day and topical ciprofloxacin 6 hourly. Culture and sensitivity result from the discharge came back as *Burkholderia pseudomallei* that resistant to cloxacillin. This case was further discussed with infective disease team. She was started with intravenous ceftazidime 2 grams tds and guttae

ceftazidime 5% qid. However, blood culture result growth no organism. Chest X-ray and ultrasound abdomen showed no evidence of abscess or systemic melioidosis disease. Patient completed fourteen days of treatment. Oral sulfamethoxazole and trimethoprim 800mg/160mg were also added. Patient responded well to treatment given. Repeat culture and sensitivity growth no organism. The fistula had sealed with no more discharge.

During followed up, probing and syringing of the lacrimal drainage system was performed. The right nasolacrimal duct was obstructed. Patient was advised to undergo acryocystorhinostomy. However, she was not keen for operation. She was then reviewed regularly in ophthalmology clinic.

4.0 Discussion

Burkholderia pseudomallei is the causative agent for disease named melioidosis. It is a gram-negative saprophyte that is abundantly found in soil and pooled waters such as rice paddy fields in tropical countries. It is considered endemic disease in Southeast Asia especially in Thailand and northern part of Australia.^{4,12} It primarily infects horses, cows, donkey and mules. It can infect human by direct traumatic inoculation (through wounds or cuts), inhalation and ingestion contaminated food or water.^{4,8,11} Adult onsets diabetic mellitus with poor blood glucose control is strongly related to melioidosis.^{6,8,11} Others conditions that are predisposed to melioidosis infection are renal disease, liver cirrhosis, thalassemia and chronic alcoholism.¹¹ Skin and subcutaneous tissue abscess formation is the second commonest presentation of melioidosis following pneumonia.^{6,8,11,12} Melioidosis septicaemia has a high mortality rate.^{6,8,9} Delayed in diagnosis, delayed admission and absence of prompt acute phase treatment contributed to the high mortality rate of melioidosis. Many countries consider melioidosis as an emerging infectious disease due to increase in plants and animals being transported around the world.^{5,10}

Burkholderia pseudomallei is not a common organism isolated in acute dacryocystitis. In fact, it was not documented in microbiology profile literatures of lacrimal abscess. There is a case report from Thailand of similar incidence, unfortunately the case was reported in Thai language and the English translated version is not available.¹³

In this case, the patient is a pre-existing adult onset diabetic mellitus, has gardening and farming as her routine work predispose her for melioidosis infestation. The most possible of pathogenesis will be direct inoculation to the eye, thus infecting the lacrimal drainage system. Bacteraemia and haematological spread was unlikely in view patient was not febrile, no systemic complaints and investigations yield no systemic involvement. The culture and sensitivity result was reliable, as the sample was collected following a sterile, normal saline dressing over the fistula wound.

Treatment for melioidosis is biphasic and of long duration, this is due to the nature of the *Burkholderia pseudomallei* infection which has a rapid progression and ability to establish latent infection due to its deep seated foci or abscesses.¹⁴ *Burkholderia pseudomallei* is well known for its antibiotic resistance and refractory to therapy.¹⁴⁻¹⁶ This is due to the unique property of the bacteria itself, such as exclusion and efflux from the cell, enzyme inactivation, altered target site, metabolic bypass and drug sequestration.¹⁴ *Burkholderia pseudomallei* is more susceptible to drugs such as imipenem, ceftazidime, co-amoxiclav, piperacillin/tazobactam and doxycycline.^{9,11,14-16} However, more case reported of emerging of ceftazidime-resistant melioidosis especially in Southeast Asia.¹⁴⁻¹⁶

Beside topical ceftazidime eye drops, patient was started with an intensive phase of intravenous ceftazidime for fourteen days, and follow by eradication phase of oral Bactrim for three months. Oral Bactrim was started one week after beginning of intravenous ceftazidime (not following completion of intravenous ceftazidime) because oral Bactrim require three to four days to achieve steady-state serum level. Patient responded well to the above treatment regime, with previous history of poor response to the oral antibiotics prescribed by the casualty department, these proved that the organism is likely to be *Burkholderia pseudomallei*.

Skin and soft tissue infection can serve as foci to predispose for disseminated melioidosis. This can eventually lead to bacteraemia and septic shock. Therefore, this patient was treated aggressively according to the standard melioidosis regime. Investigations such as blood culture to look for bacteraemia, chest imaging to look for pneumonia and abdominal ultrasound to look for deep seated abscess in liver and spleen was done during admission and repeat during follow up in clinic.

5.0 Conclusion and recommendation

Burkholderia pseudomallei should be consider as a rare cause for acute suppurative dacryocystitis in South East Asia region. Systemic investigation and imaging is important to look for deep seated abscess in various organ.

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Declaration

Author(s) declare that there is no financial interest in the above case.

Authors contribution

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