·Case Report·

Successfully treated rare presentation of orbital melioidosis

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Abstract

• AIM: To report a rare case of melioidosis presenting as orbital abscess, who was successfully treated with high dose of intravenous ceftazidime.

• METHODS: A case report.

• RESULTS: A 55-year-old Malay gentlemen who was newly diagnosed with diabetes mellitus, presented with prolonged low grade fever for three weeks and left eye swelling for five days duration. Initial CT scan of brain and orbit showed left periorbital cellulitis and acute left sphenoidal sinusitis. Initial swab culture grew Pseudomonas sp. His general condition improved with regular antibiotics. However, upon completion of intravenous therapy his condition worsened and the left eye became more proptosed. Repeat CT scan of the brain and orbit showed left eye orbital abscess with intracranial exten-sion. Swab culture from fistula of the lateral part of upper eyelid showed Burkholderia pseudomallei. He was treated with high dose of intravenous ceftazidime, oral co-trimoxazole for the acute management and on maintenance dose of oral co-trimoxazole for 2 months. He responded well to treatment and had no relapse up to one year post treatment. Unfortunately his left eye vision was not salvageable.

• CONCLUSION: This case illustrates a rare presentation of orbital abscess due to melioidosis which was complicated with cerebral abscess and septicemia. An accurate diagnosis was essential and high dose of susceptible antibiotics was important for the institution of therapy to successfully treat this potentially fatal condition.

• KEYWORDS: melioidosis; orbital abscess; cerebral abscess; high dose parenteral ceftazidime

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INTRODUCTION

Melioidosis is caused by Burkholderia peudomallei, formerly known as Pseudomonas pseudomallei, a kind of gram-negative bacilli[1]. It is endemic in Southeast Asia and Northern Australia. It has a variable form of presentation, from mild to potentially fatal infections[1-4]. The commonest presentation is pneumonia with or without septicemia but other presentation can involve soft tissue, bone, joint, genitourinary system, CNS, facial or ocular infections [1-4]. However, melioidosis presented with orbital abscess was very rarely reported[5,6].

Melioidosis poses a therapeutic challenge to the managing physicians. Inadequate management can cause deterioration of condition, and even carry mortality. In this case, we described a patient who presented with orbital abscess with temporal lobe extension due to melioidosis. He was treated with high dose antibiotics on a prolonged regime in the acute phase followed by a lower dose maintenance therapy for the eradication of the organism.

CASE REPORT

A 55-year-old Malay gentleman was newly diagnosed with diabetes mellitus during admission, presented with history of prolonged low grade fever for three weeks and left eye proptosis for five days. Initially, his left eye vision was noted to be mildly impaired. Urgent CT scan of the orbit and brain showed features of left periorbital cellulitis which was confined to the intraconal region. He developed septicemia while being admitted which was supported by the blood culture results which grew Pseudomonas sp. Patient was treated with intravenous ceftazidime 1mg twice per day, cloxacillin 500mg four times a day and metronidazole 500mg three times per day for 2 weeks. His general condition and orbital proptosis improved.

However, upon completion of the intravenous antibiotics, his general condition deteriorated and the left eye became more proptosed . Repeat CT scan of orbit and brain showed features of periorbital cellulitis with small abscesses involving the intra- and extraconal areas with extension to the left temporal lobe. MRI of brain and orbit was subsequently done, which showed similar findings (Figure 1). There was no evidence of cavenous sinus thrombosis on magnetic resonance angiography (MRA) sequences (Figure 1). In our hospital, Burkholderia pseudomallei was isolated from a swab at the fistula of the upper eyelid, which was sensitive to ceftazidime and ciprofloxacin and resistant to gentamicin. Serology test also showed presence of immunoglobulin G for Burkholderia pseudomallei. Intravenous high dose parenteral ceftazidime 2mg three times per day with oral co-trimoxazole 650mg twice per day were then started for two months.

The patient worked as a shopkeeper, with previous occupation at a sawmill when he was younger. He denied being involved in agricultural work or any previous history of eye trauma. He also denied intravenous drug abuse, contact with tuberculosis patients or sexual promiscuity.

On examination, his general condition was satisfactory and vital signs were stable. No signs of meningism were noted. Cardiovascular and respiratory systems were normal. Liver enlargement was palpable, but non-tender.

Eye examination showed no perception to light and positive reverse afferent pupillary defect over the left eye was noted, signifying the optic nerve was compromised. The left eye was proptosed with severely injected and chemotic conjunctiva (Figure 2). There was pus discharged from a fistula at the lateral part of upper eyelid (Figure 3). Cornea was clear. Anterior chamber was deep and quiet. Movements of the left eye were restricted in all directions of gaze. Optic disc was pale with presence of choroidal folds at the macula. Intraocular pressure was normal. There were mild diabetic retinopathy changes on both eyes.

Full blood picture showed leukocytosis with predominant neutrophils. Chest X-ray showed right upper lobe consolidation in accordance with previous lung infection. Mantoux test was negative and sputum was not producible

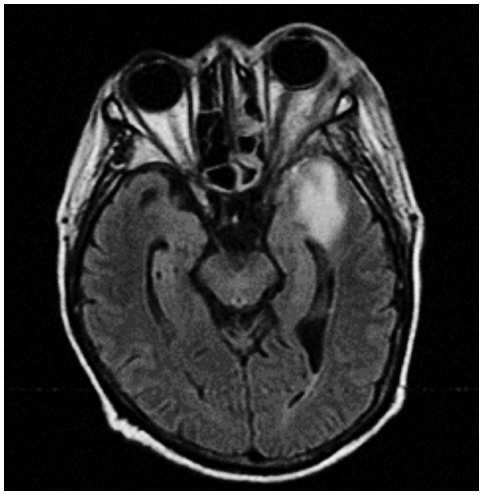

Figure 1 MRI of brain showed left periorbital cellulitis with small abscesses, intra – and extraconal fat involvement with extension to the left temporal lobe

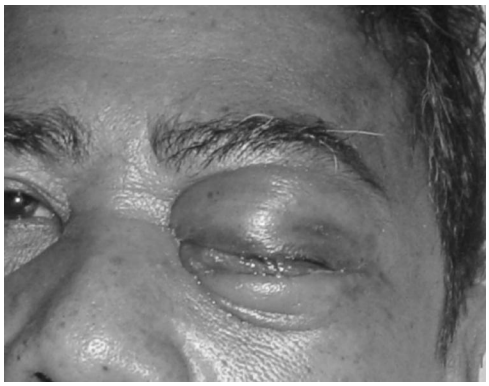

Figure 2 The left eye showed injected and chemotic conjuntiva

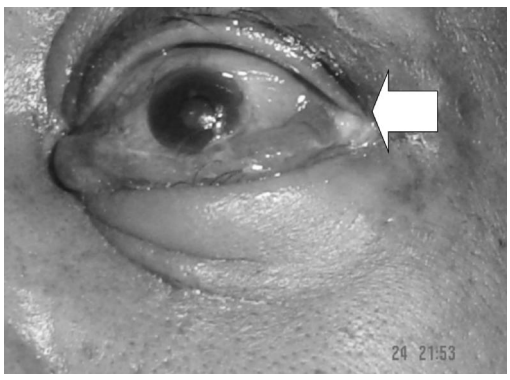

Figure 3 Pus discharged from fistula of the lateral part of upper eyelid (arrow)

for acid-fast bacilli (AFB) tests. Liver function test was elevated but ultrasound of the abdomen did not reveal any liver abscess. Random blood sugar was high which was later controlled with insulin.

His condition showed remarkable improvement after high dose parenteral ceftazidime 2mg three times per day with oral co-trimoxazole 650mg twice per day. Unfortunately, the left eye vision was not salvageable. For eradication therapy, he was planned to receive oral co-trimox-

azole for 6 months at which this patient had defaulted follow-up. However, he had showed no relapse for more than one year.

DISCUSSION

Melioidosis is caused by motile, oxidative positive, gram negative aerobic bacilli, Burkholderia pseudomallei, formerly known as Pseudomonas pseudomallei. It is endemic in Southeast Asia, Northern Australia, some parts of India and sporadic over the other parts of the world[1]. However, there were few articles on the incidence in Malaysia[2-4]. Most of the cases presented with pneumonia (ranges from 40% to 67%)[1-4]. The incidence of ocular involvement was very rare. One reported case from Malaysia was a patient who presented with orbital and parotid abscess with intracranial extension[5]. Another reported case was in 1996 from Singapore [6]. Both cases presented with orbital cellulitis in patients who had diabetis and they ended up with mortality. It is believed that many other cases were not reported or undiagnosed since it is a non-notifiable disease in Malaysia. In Singapore it has become a notifiable disease since 1996.

Mortality rate of melioidosis in Malaysia is high, 38% died within 24 hours of admission and 42.9% died after 72 hours of admission [3]. Diabetics were a susceptible group [1-4]. Most of the survivors responded with ceftazidime as a standard regime antibiotic. However, the recurrence rate is high even after 20 years. In our case, the patient's condition deteriorated after completion of intravenous ceftazidime 1mg for 2 weeks. A repeat CT scan of brain and orbit showed worsening of left orbital abscess, with extension to the left temporal lobe. A prolonged course of high dose parenteral ceftazidime 2mg three times per day plus oral co-trimoxazole 650mg twice per day were given for 2 months as acute phase treatment. The patient was planned for maintenance dose of oral co-trimoxazole 650mg twice per day for 6 months. On our last follow-up, he did not show any signs of recurrence up to one year.

In our patient, the diagnosis of melioidosis was picked up by positive culture of the blood and swab from the fistula of upper eyelid. It was also supported by the presence of Immunoglobin G to the organism.

This case highlights a patient with melioidosis presented with orbital abscess, which was a rare presentation of melioidosis. It was complicated with septicemia and cerebral abscess. There was a need to prolong and increase the dose of susceptible antibiotics in a case of melioidosis with multiple involvement of soft tissue and brain with septicemia, to prevent life threatening and vision threatening conditions.

REFERENCES

1 Cheng AC, Currie BJ. Melioidosis: epidemiology, pathophysiology, and management. *Clin Mocrobiol Rev* 2005;18(2):383–416

2 How SH, Ng KH, Jamalludin AR, Shah A, Rathor Y. Melioidosis in Pahang, Malaysia. *Med J Malaysia* 2005;60(5):606–613

3 Pagalavan L. Melioidosis: the Johor Bahru experience. *Med J Malaysia* 2005;60 (5):599–605

4 Heng BH, Goh KT, Yap EH, Loh H, Yeo M. Epidemiological surveillance of melioidosis in Singapore. *Ann Acad Med Singapore* 1998;27:478–484

5 Mohd Fauzi AR,Humairah SC,Azarisman Shah MS,Tahir M.Meliodosis presenting as orbital and parotic abscess with intracranial extension. *The International Medical Journal* 2002;1(2):www.e-imj.com

6 Wong PK,Ng PH.Melioidosis presenting with orbital cellulitis. *Singapore Med J* 1996;37(2):220–221