

The man with the boggy head: cranial melioidosis

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ABSTRACT

Melioidosis is known to cause abscesses in various organs, including the cranium, though less commonly. We present a patient with scalp abscess and subdural empyema that was visible on computed tomography of the brain. The neurosurgical drainage grew *Burkholderia pseudomallei*. Despite our best effort to treat the patient using parenteral antibiotics and surgical drainage, the patient did not survive.

Keywords: *Burkholderia pseudomallei*, cranial melioidosis, subdural empyema

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INTRODUCTION

Melioidosis is endemic to Southeast Asia, China, the Indian subcontinent and parts of Australia. The diagnosis can be elusive to the unsuspecting doctor and can often be mistaken for other maladies. Hence, the name, “the great imitator”, has been aptly coined by some clinicians.⁽¹⁾ We describe a case which was initially diagnosed as scalp and subdural haematoma complicated by dengue haemorrhagic fever, but was later confirmed to be cranial melioidosis with septicaemia.

CASE REPORT

A 48-year-old man with diabetes mellitus, who was not on regular treatment, presented with a one-month history of a painless boggy scalp swelling associated with intermittent low-grade fever and lethargy. There was no preceding head trauma. The patient had previously sought traditional treatment but found no relief. He was then admitted to a district hospital but requested for discharge the next day to seek further treatment at a private hospital.

At the private hospital, the patient was confused and jaundiced. He had scalp swelling, which the attending physician mistook to be a scalp haematoma. Blood tests showed a haemoglobin level of 11.9 g/dl, leucocyte count of 4300/ul (neutrophils 87%, lymphocytes 10%), platelet count of 17,000/ul and positive dengue IgM. Computed tomography (CT) of the brain showed a left parietal subdural haematoma with scalp swelling

overlying the parietal bones (Fig. 1). Chest radiograph and CT revealed consolidation and cavitation of both the upper lobes, with bilateral hilar lymphadenopathy and pleural effusion. The diagnosis was dengue haemorrhagic fever with subdural and scalp haematoma. Pulmonary tuberculosis and lymphoma were also considered as the differential diagnoses. The patient was transfused 12 units of platelets and started on intravenous meropenem 1 g tds empirically for suspected superimposed bacterial infection with sepsis. After being warded for five days in the private hospital, he was referred to our hospital for further management due to financial constraints.

At our hospital, the patient was still confused but was able to obey simple commands. His blood pressure was 125/90 mmHg, pulse rate 131 beats/min, temperature 38°C and oxygen saturation 91% on nasal cannula oxygen delivered at 3 l/min. There were coarse crepitations in both the lower zones of his chest. A large, soft and fluctuant swelling was felt at the top of his scalp. His renal function test was normal, and the hepatitis B and C screening tests were negative. His liver function was deranged with aspartate transaminase 161 mmol/l, alanine transferase 118 mmol/l and total bilirubin 200 umol/l (mixed hyperbilirubinaemia). The platelet count was 10,000/ul, haemoglobin 11.3 g/dl and total leucocyte count 3700/ul (predominantly neutrophils).

The patient was provisionally diagnosed with severe sepsis secondary to melioidosis and continued to receive intravenous meropenem 1 g tds. Insulin was started for blood sugar control. A referral was made for neurosurgical opinion. Two days after admission, the patient's condition did not improve despite antibiotics and supportive care. He developed severe hypoxia, and was electively intubated and ventilated. Dengue enzyme-linked immunosorbent assay (ELISA) serology was negative and blood culture confirmed the presence of *Burkholderia pseudomallei*.

On Day 7 of admission, after achieving a satisfactory platelet count for surgery, the patient underwent incision and drainage of the boggy swelling on his scalp, which removed 150 ml of pus. The pus culture also grew *Burkholderia pseudomallei*. A strong suspicion that the left parietal subdural collection was a subdural empyema led to an evacuation procedure being planned. However,

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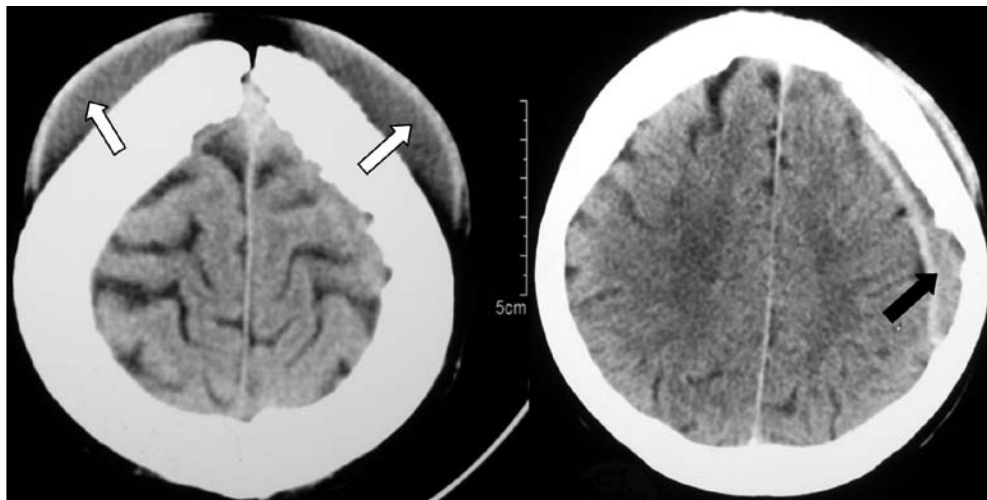


Fig. 1 Axial CT images of the brain show the scalp (white arrows) and subdural (black arrow) collections.

the patient developed non-oliguric acute renal failure and disseminated intravascular coagulopathy, which deferred further surgical intervention. He was then haemodialysed with a femoral double-lumen catheter. Additional blood products, packed cells, fresh frozen plasma and platelets were transfused, and intravenous vitamin K was administered. An ultrasonography of the abdomen did not detect any abscesses in the viscera. The patient's fever did not subside but continued to fluctuate. Intravenous meropenem was continued at an adjusted renal dose of 500 mg bd.

Two weeks later, the patient was deemed fit for surgery. A burr hole was performed, and the sanguineous fluid was drained. However, it again grew *Burkholderia pseudomallei*. The next day, the patient's condition deteriorated, and he succumbed to his illness despite attempts at resuscitation.

DISCUSSION

The most common presentation of melioidosis is pulmonary disease, and the most severe is septicaemia, which can be fatal. Melioidotic abscesses can occur in almost any organ,⁽¹⁾ such as the liver, spleen, prostate, and sometimes in the cranium. Diabetes mellitus, though not a pre-requisite, is a common predisposing factor for contracting melioidosis. Our patient had diabetes mellitus and was unfortunate to have developed melioidotic septicaemia with pulmonary and cranial involvement.

The organism, *Burkholderia pseudomallei*, a Gram-negative bacillus, grows on most culture media, such as blood agar and Ashdown's selective medium, but may be difficult to identify and differentiate from other organisms such as *Burkholderia cepacia*. The introduction of Francis medium in recent years has improved the identification of *Burkholderia pseudomallei*, and a preliminary report

can be generated within 18–24 hours.⁽²⁾ Serological tests, using the ELISA method and genetic techniques such as polymerase chain reaction, have also been used to aid in the diagnosis and monitoring of melioidosis.

Although melioidosis is commonly encountered in clinical practice in Malaysia, cranial melioidosis is unusual. In a prospective study conducted in the Royal Darwin Hospital,⁽³⁾ only 12 cases of neurological melioidosis out of a total of 232 cases of melioidosis were documented over a period of nine years. Another prospective study conducted in Thailand identified only three out of 191 melioidosis cases evaluated in 2005.⁽⁴⁾

In order to curb infection and improve patient survival, early diagnosis and the establishment of appropriate treatment are of utmost importance. Making the right diagnosis was understandably difficult in this case, as this part of the world is also endemic for dengue fever. Furthermore, the clinical picture was muddled by a false positive dengue rapid test. A report of 18 children with melioidosis in Thailand found five associated cases of dengue haemorrhagic fever, out of which three were serologically confirmed, while two were diagnosed using the World Health Organization diagnosis criteria for dengue haemorrhagic fever.⁽⁵⁾ In severe thrombocytopenia of dengue fever, spontaneous bleeding can occur anywhere. In this patient, the boggy scalp swelling, which was actually a scalp abscess, could have been easily mistaken for a scalp haematoma. However, the patient's prolonged symptoms and pulmonary disease did not concur with classical dengue fever.

The antibiotics used to treat melioidosis include ceftazidime, co-amoxiclav and cefoperazone/sulbactam plus co-trimoxazole. In severe illness, carbapenems are used. Intensive parenteral therapy can be given

for at least ten days, followed by a longer duration of eradication therapy with oral antibiotics, such as a combination of doxycycline and co-trimoxazole, which can last 12–20 weeks.⁽⁶⁾ Co-trimoxazole is substituted with co-amoxiclav in patients allergic to sulphur-containing drugs. Protracted treatment and monitoring are necessary as the disease frequently relapses.

Subdural empyema is potentially life-threatening. Although multiple abscesses in the viscera are not usually drained, surgical drainage in intracranial collections had improved survival in some cases. Primary craniotomy is the preferred method of drainage of subdural empyema⁽⁷⁾ as it allows wide exposure, adequate exploration and better evacuation. Stereotactic burr hole placement and irrigation are less desirable due to decreased exposure and possible incomplete evacuation. In a case series reported by Chadwick et al in Singapore, all five patients who had cerebral melioidosis confirmed by positive growth from neurosurgical drainage survived after surgical drainage. The patients' clinical presentations included seizures, meningitis, altered level of consciousness, limb weakness and facial palsy. These patients had cerebral abscesses (four out of five patients), subdural empyema (two out of five patients), sinusitis (four out of five patients) and vault osteomyelitis (one out of five patients).⁽⁸⁾ Our patient had undergone an early surgical drainage of the scalp abscess, but the drainage of the subdural collection was

considerably delayed, which had likely contributed to the patient's demise.

In conclusion, melioidosis can mimic other illnesses. In endemic areas, the diagnosis should be suspected especially among diabetic patients who present with sepsis. Melioidotic subdural empyema is undoubtedly life-threatening. The management includes prompt diagnosis, the administration of appropriate antibiotics and early surgical drainage.

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