Australasian Medical Journal [AMJ 2012, 5, 8, 455-458]

Cranial melioidosis with extradural extension after a fall in the bathroom

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CASE REPORT

Please cite this paper as: Naha K, Dasari S, Kusugodlu R, Prabhu M. Cranial melioidosis with extradural extension after a fall in the bathroom. AMJ 2012, 5, 8, 455-458. http://doi.org/10.21767/AMJ.2012.1374

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Abstract

A 32-year-old diabetic male, with a past history of head injury and seizures, presented with a painful swelling over his forehead present for the past three months. Cranial MRI demonstrated the presence of a scalp collection with extradural extension through a bony defect. Biopsy from the area showed caseating necrosis suggestive of tuberculosis. Although the patient failed to return for initiation of anti-tubercular therapy for the next 11 months, the swelling did not progress, and there were no constitutional symptoms. The indolent nature of the swelling prompted re-evaluation and delayed cultures of pus from the collection grew *Burkholderia pseudomallei*.

Key Words

Cranial epidural abscess, melioidosis, *Burkholderia pseudomallei*

Implications for Practice

1. What is known about such cases? *B. pseudomallei* infection is a frequently under diagnosed disease, and is known to mimic tuberculosis.

2. What is the key finding in this case report? Melioidosis should be kept in mind as a differential diagnosis of tuberculosis. Early surgical drainage of cranial melioidosis improves outcome.

3. What are the implications for future practice? All patients presenting with features of tuberculosis should be evaluated for possible melioidosis, especially in the absence of definitive evidence of tubercular infection.

Background

Burkholderia pseudomallei is a ubiquitous soil saprophyte, widely distributed in South-East Asia and Northern Australia. Human infection results in melioidosis, a disease characterised by a wide variety of clinical manifestations. Although exact figures are not available, there has been an increasing trend of cases reported from the Indian subcontinent;[1] possibly due to the availability of improved facilities for diagnosis. In their paper on the global epidemiology of melioidosis, Cheng and Currie[2] have classified India as a country with sporadic cases of melioidosis only. However we have noted a steady increase in cases of melioidosis at our hospital, perhaps reaching endemic proportions. Unfortunately, melioidosis is not yet a notifiable disease in India, so that accurate national estimates are not available unlike Australia and Thailand.

Morphologically, *B. pseudomallei* is a gram-negative bacillus with a characteristic bipolar appearance on staining that yields a safety pin appearance.[3] It is an exceptionally hardy organism, capable of surviving in extreme environments for extended periods of time.[4]

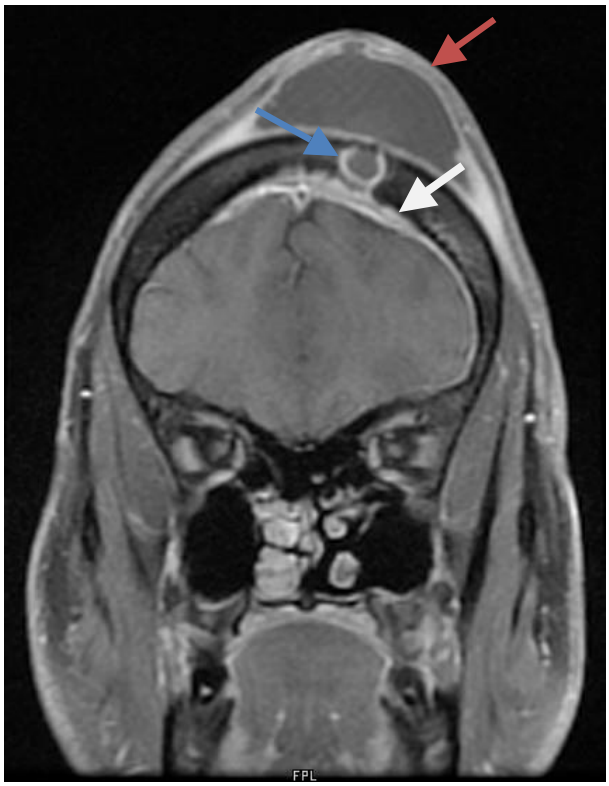
While relatively easy to isolate in culture, it can be difficult to differentiate *B. pseudomallei* from closely related non-pathogenic organisms like *B. thailandensis*. This aspect of the bacterium, along with its ability to produce chronic disseminated disease mimicking tuberculosis,[5] can frequently result in misdiagnosis, especially in countries like India with a relatively high prevalence of tuberculosis. This propensity of *B. pseudomallei* to mimic tuberculosis is exemplified in our case report. We have also highlighted the benefit of early surgical drainage on the natural history of intracranial melioidosis.

Case details

A 32-year-old male, a diagnosed diabetic and on medical nutrition therapy since the past two years, presented to our outpatient clinic with a painful swelling over his forehead. The swelling had been progressively increasing in size over the past three months, and had partially ruptured two days before his arrival to our clinic. Five months earlier, he had suffered a head injury after a fall in the bathroom, at which

time he had sustained a minor laceration involving the same portion of his forehead. The fall was followed immediately by a single episode of generalised tonic-clonic seizure lasting around one minute. He had not sought any medical help at that time, and had had no further seizures since then. He denied any constitutional symptoms such as fever or weight loss. There was no history of substance abuse.

Figure 1: Contrast enhanced MRI brain (T1 sequence, coronal section) showing a well-defined fluid intensity lesion measuring 6.7x2.7x5.7 cm in the scalp in the left high fronto-parietal region (red arrow) with an iso-intense periphery, communicating with a similar signal intensity extradural collection (white arrow) in the left frontal region through an irregular 1 cm wide calvarial defect (blue arrow).

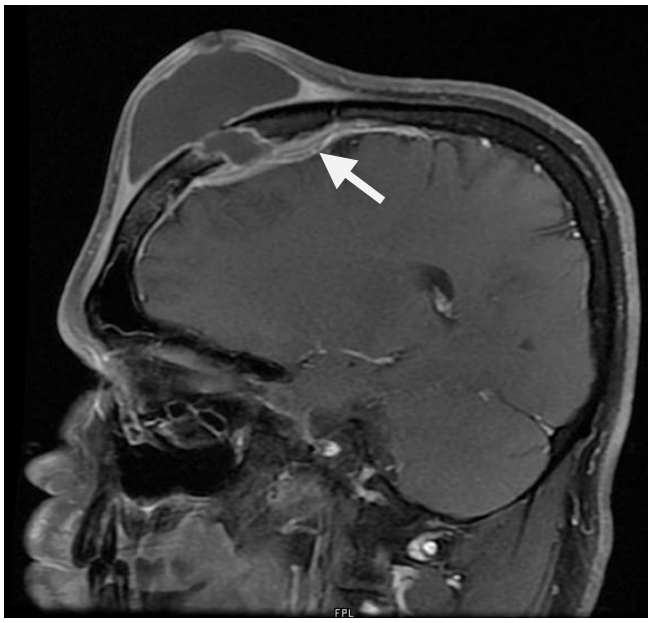


Local examination revealed a tender, warm, cystic midline swelling with sero-purulent discharge on his forehead. Neurologic examination did not reveal any focal deficits. Review of cardiovascular, respiratory and gastro-intestinal systems was essentially normal.

Routine laboratory parameters including ESR (10 mm/hr) were unremarkable except for uncontrolled hyperglycaemia with glycosylated haemoglobin (HbA_{1c}) levels at 9.0%. Cranial MRI confirmed the presence of a scalp collection (Figures 1, 2). The collection was seen to extend through a bony defect in the calvarium into the extradural space with hyperaemia over the adjacent meninges. A left frontal craniotomy was performed under general anaesthesia and the granulation tissue biopsied. Histopathologic examination showed dense fibrocollageneous tissue with infiltration by neutrophils forming microabscesses, surrounded by lymphocytes, histiocytes and areas of caseous necrosis with few palisading epithelioid cells, and dead bony spicules. Although staining for acid-fast bacilli was negative, tuberculosis was suspected based on the characteristic picture of caseating necrosis, the presence of diabetes – a known risk factor for tuberculosis, and the chronic nature of infection. Mantoux test performed was indeterminate with an induration of eight mm. Plain chest X-ray performed was normal, and revealed no stigmata of current or past tubercular infection. Initial culture reports of the pus were sterile. Although the clinical and histopathologic picture was compatible with a tubercular cold abscess, the absence of definitive evidence in the form of absence of acid fast bacilli in the biopsy, absence of radiographic signs of tuberculosis and indeterminate tuberculin skin test, prompted us to await the mycobacterial culture report. The patient was therefore treated with subcutaneous insulin and discharged with instructions to return after one month for follow-up of mycobacterial culture of the biopsy sample. In the event that the culture was positive, we planned to initiate antitubercular drugs in accordance with the DOTS regimen recommended in India. Unfortunately, the patient was subsequently lost for follow-up, returning only after another 11 months. In the interval, the swelling had persisted, now associated with a discharging sinus. Although he had not yet received anti-tubercular therapy in any form, he appeared relatively well. Systemic symptoms were once more conspicuous by their absence. The remarkable indolent nature of the swelling prompted us to review his previous medical records, which revealed that delayed cultures of pus and granulation tissue had grown *B. pseudomallei*, with a typical sensitivity to co-amoxiclav, ceftazidime, cotrimoxazole and meropenem. A repeat biopsy of the discharging sinus was performed and showed no evidence of tuberculosis. Moreover, mycobacterial culture of granulation tissue from the first biopsy remained sterile.

Having definitively ruled out tuberculosis, pharmacotherapy for melioidosis was initiated with parenteral co-amoxiclav (1.2 g IV q8h) for 14 days, followed by maintenance phase with oral cotrimoxazole (320/1600 mg PO q12h) for 20 weeks. The patient made a rapid recovery with complete resolution of the discharging sinus and the underlying abscess. After more than a year of follow-up, the patient is healthy with no recurrences.

Figure 2: Contrast enhanced MRI brain (T1 sequence, sagittal section) showing the same lesion. Note the contiguous meningeal enhancement in the frontal region (white arrow)



Discussion

There are several points of interest in our case report. Risk factors for *B. pseudomallei* infection including chronic pulmonary and renal disease, occupational exposure to contaminated water sources and excessive alcohol consumption are often cited as an important clue in making a diagnosis of melioidosis. While such risk factors are definitely helpful, their absence does not rule out a possibility of melioidosis. In our patient for instance, diabetes mellitus was the only identifiable risk factor. Although diabetes mellitus is a well-known risk factor for melioidosis,[6-7] generally attributed to impaired neutrophil function, it is also associated with a wide range of other infectious diseases. Therefore, it is important to consider melioidosis as a differential diagnosis in any patient presenting with a compatible clinical picture regardless of the presence or absence of risk factors.

Our case also represents a rare manifestation of melioidosis. Among the various clinical presentations of *B. pseudomallei*, pulmonary infection and multiple visceral abscesses are frequently encountered. In contrast, neurological involvement is unusual in melioidosis. A seminal study performed in northern Australia[8] identified only 14 cases of neurologic disease in 540 cases over 20 years, while a similar study in Thailand identified only three cases out of 190.[9] Vachvanichsanong and colleagues reviewed paediatric cases with neurologic melioidosis and found only eight such cases.[10] Furthermore, a proportion of these cases had cerebral abscesses as part of disseminated systemic infection. An Indian study by Kumar et al,[11] reported six cases of cranial melioidosis. However, in all these cases, there was either isolated involvement of brain parenchyma in the form of intra-cerebral abscesses or osteomyelitis without extension into the extradural space. Cranial disease produced by local infection or inoculation has been reported in only a handful of studies, including a case series by Chadwick et al[12] and a single case report by Kuan et al.[13] Interestingly, most of these cases displayed features of severe sepsis. Indeed, the patient reported by Kuan and colleagues[13] received prolonged therapy with intravenous meropenem, but succumbed to the disease nonetheless. In contrast, our patient remained relatively asymptomatic despite receiving no treatment for over 11 months. Notably, drainage of the abscess was performed early in our case, when compared to the case described by Kuan. Here surgical drainage of the scalp collection was performed after 12 days of intravenous antibiotic therapy, and intracranial drainage via burr hole was attempted after another fortnight. This suggests that early drainage of such abscesses may play a major role in influencing patient outcomes, perhaps comparable to antibiotic therapy itself. Further study is required to elucidate this possibility.

This case report demonstrates an unusual presentation of melioidosis, highlighting its potential for mimicking tuberculosis both clinically as well as pathologically. It also emphasises the importance of early drainage in such cases, potentially delaying the natural history of the disease, and preventing significant morbidity even in the absence of appropriate antibiotic therapy. The relatively asymptomatic condition of the patient is also noteworthy, especially in the context of an extradural empyema, and is in sharp contrast to previous reports of intracranial melioidosis, where patients displayed significant morbidity and mortality.

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ACKNOWLEDGEMENTS

None.

PEER REVIEW

Not commissioned. Externally peer reviewed.

CONFLICTS OF INTEREST

The authors declare that they have no competing interests.

FUNDING

None.

PATIENT CONSENT

The authors declare that:

1. They have obtained written, informed consent for the publication of the details relating to the patient in this report.
2. All possible steps have been taken to safeguard the identity of the patient.
3. This submission is compliant with the requirements of local research ethics committees.