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Case Report

Massive calvarial melioidosis abscess following minor trauma in rural areas of Thailand

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ABSTRACT

Background: Melioidosis is uncommon but endemic in Southeast Asia and parts of Northern Australia. Cerebral melioidosis is rare but can be spread through several routes, such as hematogenous spreading or the direct inoculation of organisms from wound contamination with soil. It can cause devastating sequelae if the treatment is delayed. However, with early and adequate treatment, patients can recover and have a good quality of life.

Case Description: A 62-year-old diabetic male presented with epilepsy 2 months after a head injury. A computed tomography scan revealed an abscess extending from the subgaleal layer to the subdural with osteomyelitis. A craniotomy was performed to remove the abscess. Melioidosis was identified from pus culture. Intravenous meropenem with Bactrim was started, followed by oral doxycycline and bactrim. The patient recovered with no seizure episodes. This patient showed a rare but straightforward infection from direct inoculation in a wound contaminated with soil. Incubation time could be up to 2 months. The infection originates from previously lacerated scalp tissue and invades the skull, causing osteomyelitis and epidural abscess. Prompt treatment brings a good outcome. In patients with risk factors and a suspicious history, broad-spectrum antibiotics should be initiated after removal of the abscess.

Conclusion: Melioidosis is still endemic in Thailand. Doctors should be aware of this organism in patients with high-risk factors or travelers who have just returned from an endemic area. Patients should be treated early with an adequate dose and duration of anti-melioidosis.

Keywords: Atypical, Brain abscess, Immunocompromise, Infection, Melioidosis, Rural

INTRODUCTION

Melioidosis is an infection caused by *Burkholderia pseudomallei*, a non-spore-forming Gram-negative bacillus.[27] This infection can be found in water and moist clay soil, especially in rural tropical areas.[1,27] It is endemic in Southeast Asia and parts of Northern Australia,[8] where the organism can be found growing in rice fields, predominantly in Northeastern Thailand.[27] Melioidosis can be found in Western countries such as the United States of America; however, most people become infected when traveling to tropical areas rather than by local pathogens.[1] Commonly, melioidosis can form an abscess in the lungs, liver, spleen, and skeleton muscles, leading to severe infection or septicemia. Melioidosis can be fatal due to its resistance to antibiotics.

Before the era of Anthrax, *B. pseudomallei* was considered to be a bioweapon due to its high fatality rate.[3] However, due to advancements in antibiotic innovation, melioidosis now responds to some antibiotics, and the infection can be eradicated effectively.

Some predisposing factors significantly related to melioidosis infection are stated in recent publications. Diabetes mellitus, heavy alcohol consumption, pre-existing renal disease, thalassemia, and occupational exposure are all confirmed to be significant risk factors for melioidosis and melioid septicemia.[2,26] Immunocompromised populations should be made aware of melioidosis and the important role played by soil and water contamination.[26,27] Furthermore, a relationship exists between melioidosis cases and the amount of rainfall.[2] Therefore, those living in rural tropical areas are at increased risk of melioidosis infection.

Most of the infections are transmitted by direct inoculation from wet soil, water, or the inhalation of aerosol particles.[2,6,10] A small scratch or open wound contaminated with soil and water can introduce the organism into a person's body. Rarely this disease can spread through the perinatal or zoonotic route.[2,23] There are no definite conclusions as to which type of transmission leads to severe septicemia. Moreover, the immunocompromised status of the host, such as poor blood sugar control or severe renal failure, tends to play a more crucial role than the route of transmission.

The involvement of melioidosis in the central nervous system is rare, as demonstrated by the paucity of reports.[2,25] Presenting symptoms usually consist of fever and an alteration in consciousness.[11] Different routes of transmission can cause cerebral involvement. First, the hematologic spreading of systemic melioidosis causes intraparenchymal and brainstem abscesses, resulting in cranial nerve palsy.[3-5,14,15,25] Second, osteomyelitis of the skull and extra-axial abscess also present with a headache and scalp swelling.[12,13,16,22] The latter mostly results from direct contact with bacteria from a wound contaminated with dirty water and soil, especially in an immunocompromised host. The majority of cases are found in endemic areas, some involving travelers from outside the region.[7,11,20]

This report shows the rare presentation of osteomyelitis and extended to form subdural abscesses caused by melioidosis infection and how they can be successfully treated.

CASE PRESENTATION

A 62-year-old Thai male with poorly controlled type II diabetes and ischemic heart disease had a motorcycle accident. He briefly lost consciousness and suffered an abrasion wound and contusion at the right frontal forehead. On awakening, he experienced no nausea or vomiting. He did not go to the hospital after the accident or receive any antibiotic treatment.

Two months later, he developed a generalized tonic-clonic seizure lasting for 5 min with jerky movement of the eyes. After that event, he regained consciousness but had persistent left hemiparesis. He was treated with traditional medicine, but his condition did not improve. Before admission, his seizures became more frequent, occurring up to 7–8 times/day with 5 min duration each time. Three months after the accident, he still experienced the same seizure pattern 4 times a day. He had post-ictal drowsiness, and his relative finally brought him to the hospital.

The patient was alert and responded to all stimulation at the hospital. Seizure activity was not present at that time. On examination, a bulging mass was revealed at the frontal forehead, fluctuating but not tender, with no sign of inflammation. All cranial nerves functioned normally. No motor or sensory weakness was detected.

Laboratory investigations revealed white blood cells 8,260 cells/cu mm^3 with 53.4% neutrophils. The blood sugar level was 280 mg/dL, while electrolytes and kidney function were within normal limits. The contrast-enhanced axial computed tomography (CT) scan revealed a rim-enhancing subgaleal fluid collection in the right frontal region and a subdural fluid collection in the adjacent right frontal convexity [Figure 1]. No bone destruction was present at the first scan. The contrast-enhanced magnetic resonance imaging (MRI) was performed 3 days later [Figure 2]. The finding depicted a typical appearance of subgaleal and subdural abscesses, showing a rim-enhancing cystic lesion with restricted diffusion. Diffuse dural enhancement was more prominent in the right cerebral convexity. The right frontal bone exhibited bone marrow edema and enhancement, suggestive of acute osteomyelitis. Intravenous Keppra infusion was given immediately. A craniotomy was performed in an urgent setting. Yellowish pus was found under the scalp tissue, along with osteolytic invasion of the skull and epidural abscess. Copious irrigation was conducted to remove and clean the abscess. The pus was sent for culture, and carbapenem was started as a broad-spectrum antibiotic for brain abscess. Three days later, *B. pseudomallei* was found in the pus culture. This showed good sensitivity to ceftazidime and bactrim. There was no other organ involvement shown in the CT scan of the chest and abdomen. No bacteremia was found in the hemoculture. Meropenem was continued, and bactrim injection started for 4 weeks in the active phase, then switched to oral doxycycline 200 mg/day with co-trimoxazole (80/400) consisting of trimethoprim 640 mg/day and sulfamethoxazole 3200 mg/day for 6 months. We followed the patient with a clinical and follow-up CT scan of the brain after the complete regimen.

After treatment, the patient no longer experiences seizures. No new neurodeficit was found after surgery. His endocrinologist tightly controlled his blood sugar. Follow-up imaging showed a resolved lesion without recurrent abscess [Figure 3].

DISCUSSION

Melioidosis is still endemic in Southeast Asia and parts of Northern Australia.[1,2,4,6] Central nervous system involvement might be overlooked due to its rarity. However,

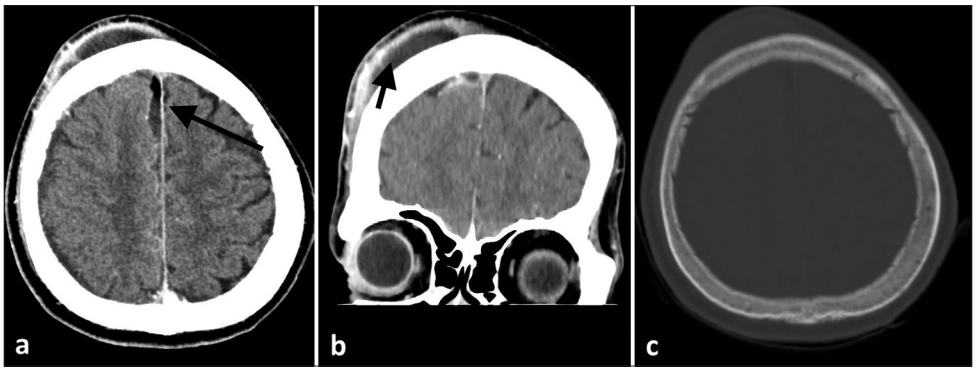

Figure 1: (a) The contrast-enhanced axial CT scan revealing a subdural fluid collection in the adjacent right frontal convexity (Long arrow), (b) A rim-enhancing subgaleal fluid collection in the right frontal region (Short arrow), (c) No bone destruction present at the first scan.

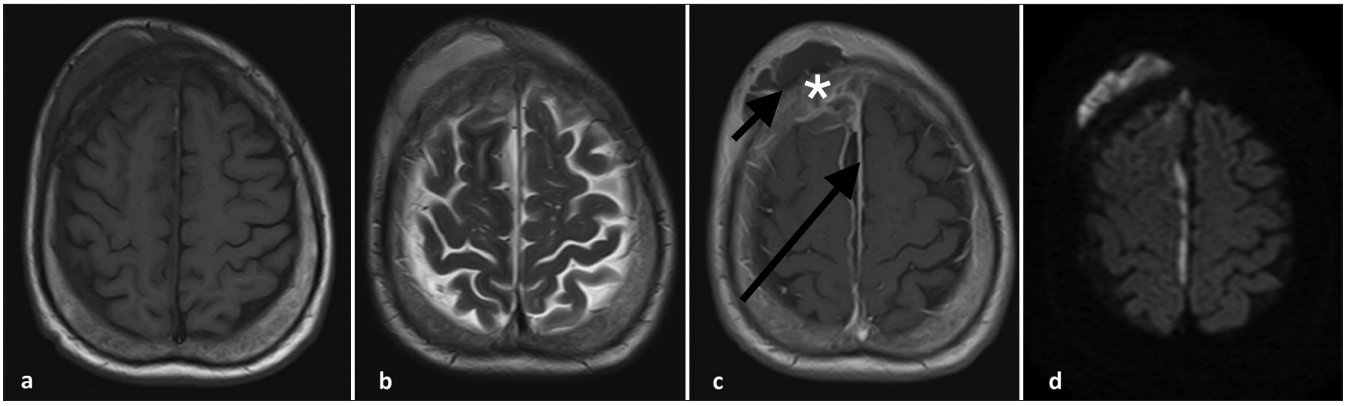

Figure 2: Contrast-enhanced magnetic resonance imaging study. (a) Axial T1-weighted image, (b) T2-weighted image, (c) Gadolinium-enhanced T1-weighted image, (d) diffusion-weighted image. Figure 2c depicted a typical appearance of a subgaleal abscess (short arrow) and a subdural abscess (long arrow), showing a rim-enhancing, cystic lesion with restricted diffusion. Diffuse dural enhancement is shown to be more prominent in the right cerebral convexity. The right frontal bone exhibits bone marrow edema and enhancement, suggestive of acute osteomyelitis (asterisk).

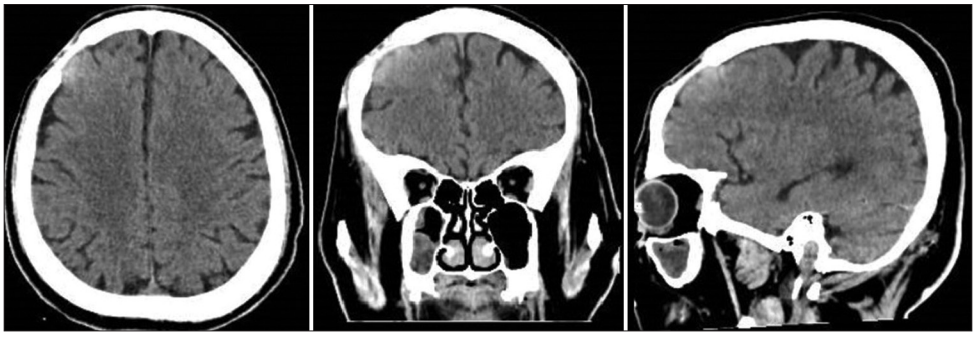

Figure 3: Resolved lesion after successfully treated. No residual abscess found.

in endemic areas, some patients with an atypical brain abscess at risk of melioidosis infection should be carefully investigated. This case report shows the uncommon but interesting presentation of cerebral melioidosis.

Cerebral melioidosis

In similarity to other bacterial brain abscesses, the most common presentation of cerebral melioidosis is fever with an alteration in consciousness. Seizure as a presenting symptom is not common in this disease but is still one of the symptoms of a cerebral abscess and may be associated with cerebral melioidosis, according to some literature.[24] Bulging at the scalp area may also mislead the diagnosis away from infection since it shows no obvious signs of inflammation, potentially delaying proper diagnosis and increasing the risk of fatality.[2,25] In the patient under study, by considering their history and presentation, it was possible to ascertain that the predisposing factors for melioidosis were poorly controlled diabetes mellitus and contamination with dirty soil from the accident scene. The laceration wound from the initial trauma caused contagious spreading with the location of an abscess in the subcutaneous area of the scalp, and the patient developed osteomyelitis from direct inoculation. For patients from endemic areas with promising health status and events that increase the risk of dirty soil and water contamination, melioidosis infection should be borne in mind. A long incubation period is possible. The patient under study experienced a 2-month incubation period, and his infection was considered subacute to chronic, which might explain the absence of inflammation signs. Patients with risk factors should be treated promptly, even though the clinical course may not be in an acute setting.

Imaging of cerebral melioidosis

Proper imaging is crucial in melioidosis. A plain radiograph of the skull may provide less information for diagnosis. A CT scan provides better imaging in this case due to its availability, good sensitivity, and specificity. In general, the findings of a CT scan can range from normal, focal brain swelling, and abscess formation depending on the stage of the disease;[5,18] thus, an MRI is more sensitive, especially in detecting early disease, although it may not be available at every hospital.

The common MRI findings are brain edema, rim-enhancing abscess, and leptomeningeal enhancement, most commonly affecting the brainstem, frontal lobe, and parietal lobe.[5,28] Some authors have reported that lesions tend to spread along the commissural and projection white matter tracts.[5,9,17] Facial and trigeminal nerves are the most common cranial nerves affected, showing thickening and enhancement of the nerve to its nucleus.[9,28] Calvaria osteomyelitis, scalp abscess, and extra-axial abscess are also reported to develop from direct infection after head injury. Spinal diseases caused by *B. pseudomallei* are very rare and can appear, such as myelitis, epidural abscess, spondylodiscitis, and paravertebral abscess.

Investigation

There are multiple ways to detect melioidosis, such as a biological test, serological test, or polymerase chain reaction (PCR) from the abscess or tissue.[19] However, culture from the blood or abscess is the most commonly used method due to the availability of the test. Some tests, such as immunoassay or PCR, may not be available in every endemic area. In the case under study, the organism was detected from the abscess culture, which is inexpensive and available at the hospital. It might take around 2–3 days to receive the result back. Meanwhile, a broad-spectrum antibiotic can be started and adjusted after the culture result has been returned.

Management

Every cerebral abscess should be eradicated in an acute hospital setting.[2] In some deep-seated intraparenchymal cases, draining could cause fatal complications or profound neuro deficits, so organism identification may be performed, followed by systemic anti-melioidosis. However, in surgically manageable cases such as osteomyelitis of the skull and epidural abscess, the pus should be promptly evacuated. An urgent craniotomy with abscess removal was performed in the patient under study due to the abscess being in an accessible location, and an antibiotic was then started. Previously, tetracycline, chloramphenicol, and co-trimoxazole were standard medications for cerebral melioidosis.[21] However, nowadays, treatments have developed considerably. Treatment steps include intensive intravenous therapy with a choice of meropenem or ceftazidime for 4–8 weeks plus co-trimoxazole orally in severe cases, then switching to oral co-trimoxazole for 3–6 months as a first-line drug. Doxycycline serves as a second-line treatment for those allergic or resistant to co-trimoxazole, with Augmentin being the third-line therapy to eradicate this infection.[2] Although relapse has been reported in several studies, after treatment, the patient in this present case recovered completely with no recurrent seizures.

Factors facilitating a good outcome in this patient consisted of the prompt management of abscess eradication and adequate antibiotics. It is reasonable to start broad-spectrum antibiotics and then tailor the treatment to specific antibiotics that show sensitivity to this pathogen. Due to the long duration of treatment, patients may not cooperate well. Communication with care providers or patients' relatives is mandatory to avoid incomplete treatment. Nowadays, melioid brain abscess responds well to treatment, and the prognosis is good. Surgeons should be aware of this disease, especially in patients with risk factors.

CONCLUSION

Cerebral melioidosis can still be found in endemic areas. It should be borne in mind for patients with risk factors such as diabetes mellitus or chronic disease and a history of open wounds contaminated by soil, prompt treatment is required to ensure good results. Emergency draining of the abscess followed by adequate antibiotics still yields an excellent outcome.

Ethical Approval

The Institutional Review Board approval is not required.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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