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Case report

Focal pachymeningitis in a returning traveler: Don't forget melioidosis

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ABSTRACT

Background: Melioidosis is an endemic disease in South-East Asia and Northern Australia caused by a Gram-negative bacillus, *Burkholderia pseudomallei*. Manifestations are wide and neurological involvement have rarely been described.

Methods: In this paper, we report a patient returning from Asia with an unusual infection including CNS involvement consistent with a melioidosis.

Results: This diagnosis was challenging and complex to carry out with multiple considerations, mainly because of the atypical nature of the germ. *Burkholderia pseudomallei* can be easily misidentified with *Burkholderia thailandensis* (rarely pathogenic to humans) during bacterial culture because of their phylogenetic proximity. The main pitfall of the management was that the responsible infectious agent was not referenced in the MALDI-TOF (considered as a bioterrorism agent) and led to a wrong strategy.

Conclusions: This case of melioidosis shows the difficulty regarding the diagnosis of this disease in a patient returning from an endemic zone and its frequent multiple organs involvement. Melioidosis is an emerging, potentially fatal disease which requires prolonged antibiotic treatment. Difficulties in clinical microbiology laboratories diagnosis of melioidosis, especially in non-endemic areas where clinical suspicion is low, may delay treatment and affect disease outcomes.

A patient in their 50 s was admitted to the emergency department for fever, headache and cough. The medical history included type 2 insulin-dependent diabetes mellitus. The patient worked as a site foreman and had recently been traveling in Thailand on the island of Koh Samui. The patient presented with high fever (40.2 °C) without sign of severe sepsis. Oxygen saturation was 98 % on room air. The pulmonary auscultation identified crackles on the left lung base. White blood cell count showed a normal count of white blood cells at 4.1 G/L (4−10) but an inflammatory state as the CRP reached 154 mg/L (< 5). A pair of blood cultures (aerobic and anaerobic bottle) was collected. The chest x-ray was consistent with left basilar pneumonia. The brain computed tomography scan (CT-scan) was normal. Empiric treatment with amoxicillin was therefore initiated for the treatment of a community-acquired pulmonary infection. The patient was hospitalized. Direct testing of blood culture revealed gram-negative aerobic bacillus. Culture on blood agar showed colonies with bipolar staining. Analysis of the organism by the Matrix-assisted laser desorption ionization time-of-flight mass spectrometry (MALDI-TOF) was in favor of Burkholderia thailandensis. According to the antibiotic susceptibility, Amoxicilline was switched to oral levofloxacin for a further 10 days allowing the patient to be discharged and return home. Five days later the patient was hospitalized again because of the persistent headaches and for abnormal movements in favor of myoclonus involving the right body side with secondary generalization and postictal state. Clobazam and levetiracetam was initiated. Analysis of cerebrospinal fluid (CSF) revealed 5 white blood cells, hyperproteinoachia (1.13 g/L), normal glycorrhachia, with negative CSF culture and negative 16S rRNA PCR. The electroencephalogram was normal. Brain magnetic resonance imaging (MRI) revealed a left frontoparietal pachymeningitis with contrast enhancing (Fig. 1A).

We were faced with a bacterial infection with multi-systemic involvement (pulmonary, neuro-meningeal), uncontrolled despite the antibiotics initiated occurring in a context of return from a foreign country (Southeast Asia). Given to the persistence of the infectious syndrome and the unusual characteristics of the initial organism

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(Burkholdelia), the microbiological diagnosis was reconsidered and the strain sent to a reference center and tested using a special bank of bioterrorism organisms. The final identification of the strain (multiplex PCR assay) was Burkholderia pseudomallei (Fig. 1B) allowing the diagnosis of melioidosis. After antibiogram analysis, meropenem was initiated (six grams per day) and continued for 3 weeks before an oral switch to trimethoprim-sulfamethoxazole for a further 10 weeks. The patient was treated overall 12 weeks. Three months after, the patient remained without symptom and brain MRI with gadolinium showed complete remission of the focal pachymeningitis.

Melioidosis is an endemic disease in South-East Asia (notably in Thailand) and Northern Australia caused by Burkholderia pseudomallei [1]. This gram-negative bacillus is acquired mainly by percutaneous inoculation and inhalation, mostly at the contact of wet soil and standing water. Melioidosis manifestations are wide: latent infection, localized cutaneous or visceral abscesses that can be acute or chronic, pulmonary infection (the most frequent) and bacteremia [2]. Fulminating forms are described and the mortality in these cases in endemic areas is estimated of 40% despite well conducted treatment [1]. Risk factors include diabetes mellitus and chronic lung disease, followed by chronic renal disease, immunosuppression, chronic alcoholism and thalassemia [3]. Neurological manifestations of melioidosis are rare and represent 5 % of cases, mainly represented by meningitis, abscesses, cranial nerve palsies and encephalitis, with a frequent involvement of the brainstem [4]. Association with pneumonia is common. An increasing number of imported cases in Europe are described associated to the development of worldwide tourism [5]. Treatment of melioidosis include an acute phase with intravenous administration for at least 10 days of ceftazidime in case of uncomplicated melioidosis. The second phase of treatment is an eradication phase using preferentially trimethoprim-sulfamethoxazole (TMP / SMX). For CNS infection, treatment is a minimum of 14 days with meropenem and 8-weeks with TMP / SMX.

We herein report a case melioidosis revealed by the association of pneumonia with bacteremia and an unusual neurological involvement (focal pachymeningitis) due to Burkholderia pseudomallei. This case of melioidosis shows the difficulty regarding the diagnosis of this disease in a patient returning from an endemic zone and its frequent multiple organs involvement. Burkholderia pseudomallei can be easily misidentified with Burkholderia thailandensis (rarely pathogenic to humans) during bacterial culture because of their phylogenetic proximity [6]. Melioidosis is an emerging, potentially fatal disease which requires prolonged antibiotic treatment. Difficulties in clinical microbiology laboratories diagnosis of melioidosis, especially in non-endemic areas where clinical suspicion is low, may delay treatment and affect disease outcomes.

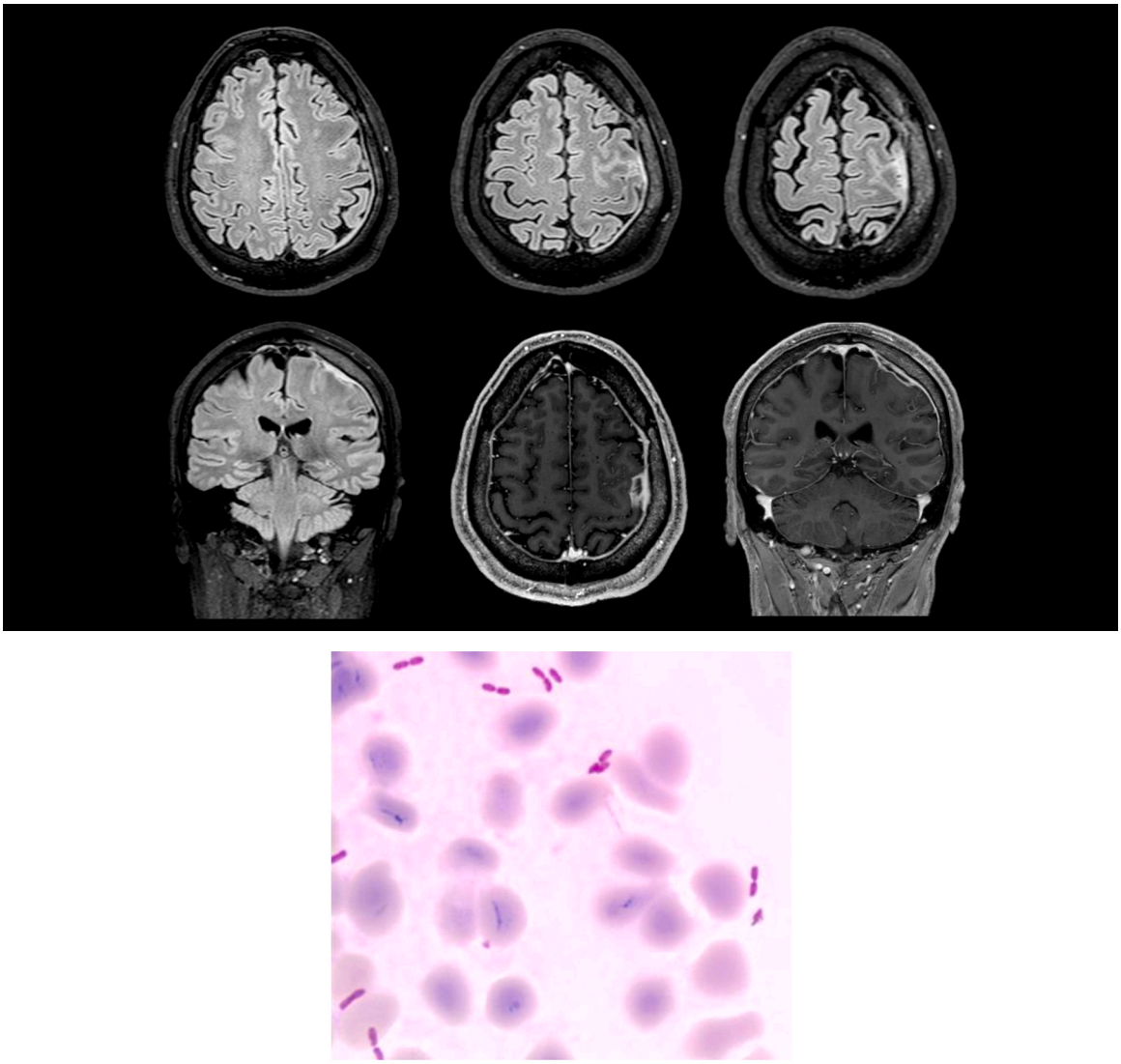

Fig. 1. A. Cerebral MRI. B. Direct testing of blood culture.

Consent

Informed consent was obtained from the patient for being included in the study.

Ethical approval

Obtained (Comité d'éthique du centre hospitalier du Havre).

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CRediT authorship contribution statement

Anne Vandendriessche, analysis or interpretation of data, acquisition of data. Franck Labbé, analysis or interpretation of data, acquisition of data. Vincent Langlois, drafting the manuscript, study concept or design.

Conflict of Interest

Authors have nothing to disclose.

References

[1] White NJ. Melioidosis. Lancet 2003;36(9370):1715–22. May 17.
[2] Birnie E, Virk HS, Savelkoel J, Spijker R, Bertherat E, Dance DAB, Limmathurotsakul D, et al. Global burden of melioidosis in 2015: a systematic review and data synthesis. Lancet Infect Dis 2019;19(8):892–902 (Aug).
[3] Suputtamongkol Y, Chaowagul W, Chetchotisakd P, Lertpatanasuwun N, Intaranongpai S, Ruchutrakool T, et al. Risk factors for melioidosis and bacteremic melioidosis. Clin Infect Dis Publ Infect Dis Soc Am 1999;29:408–13 (Aug).
[4] Currie BJ, Fisher DA, Howard DM, Burrow JN. Neurological melioidosis. Acta Trop 2000;74:145–51 (Feb).
[5] Cheng AC, Currie BJ. Melioidosis: epidemiology, pathophysiology, and management. Clin Microbiol Rev 2005;18:383–416 (Apr).
[6] Wongsuvan G, Hantrakun V, Teparrukkul P, Imwong M, West TE, et al. Sensitivity and specificity of a lateral flow immunoassay (LFI) in serum samples for diagnosis of melioidosis. Trans R Soc Trop Med Hyg 2018;112(12):568–70. Dec 1.