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Case Report

A fatal case of melioidosis with pancytopenia in a traveler from Indonesia

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ABSTRACT

Melioidosis, an infectious disease with high mortality, caused by *Burkholderia pseudomallei,* is endemic in southeast Asia and northern Australia. In Indonesia, autochthonous cases have been rarely reported, with most cases being sporadic and occurring in travelers. Herein, we report a fatal case of neurological melioidosis in a traveler from Indonesia presenting with septic shock.

1. Introduction

Melioidosis, an infectious disease with high mortality, caused by *Burkholderia pseudomallei,* a gram-negative facultative intracellular bacillus, is endemic in southeast Asia and northern Australia [1]. In northeast Thailand, *B. pseudomallei* has been reported to represent a major pathogen in community-acquired pneumonia [2]. Moreover, a limited number of autochthonous melioidosis cases have been reported in Indonesia, although these mainly occurred before 1960 [3]. Recently, most cases in Indonesia have been sporadic cases reported mainly in travelers [3]. In a previous study of acute and potentially life-threatening tropical diseases among travelers, most fatal cases were falciparum malaria, while melioidosis and severe dengue accounted for a subset of deaths [4]. Herein, we report a fatal case of melioidosis presenting with septic shock in a traveler from Indonesia.

2. Case report

A 40-year-old Indonesian man was transferred to our hospital with fever, chills, productive cough, and dyspnea. He had been in his usual health until 2 weeks before admission, while still in Indonesia, when fever and epigastralgia developed, and had subsequently been administered a proton pump inhibitor. He had not been in contact with any birds in Indonesia and had never traveled to the Middle East. One week before admission, he had arrived in Japan for a training course, after which time his symptoms had been improved for a few days. However, three days before admission, chills and abdominal pain had developed. On the day of admission, he experienced fever, chills, cough, and sputum, and visited another hospital. At that hospital, the rapid test for influenza A virus was positive, and his blood examination revealed pancytopenia. Consequently, he was transferred to our hospital for further evaluation.

On examination, his body temperature was 37.8 °C; pulse, 134 beats/minute; blood pressure, 93/62 mmHg; respiratory rate, 37 breaths/minute; and oxygen saturation, 93% in ambient air. Initial laboratory evaluation showed pancytopenia, elevated creatinine kinase, elevated aspartate transaminase, and hyponatremia (Table 1). Chest radiography revealed consolidation in the right

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Table 1
Initial laboratory evaluation.

White blood cells (×1000/μL)	1.42
Neutrophils (%)	76.8
Lymphocytes (%)	21.1
Monocytes (%)	1.4
Eosinophils (%)	0.7
Hemoglobin (g/dL)	6.6
Hematocrit (%)	19.2
Platelets (×10,000/μL)	0.9
Sodium (mmol/L)	132
Potassium (mmol/L)	3.4
Albumin (g/dL)	1.4
Total bilirubin (mg/dL)	1.3
Blood urea nitrogen (mg/dL)	16.1
Serum creatinine (mg/dL)	1.07
Aspartate aminotransferase (units/L)	120
Alanine aminotransferase (units/L)	34
Lactate dehydrogenase (units/L)	296
Creatine kinase (units/L)	1861
C-reactive protein (mg/dL)	23.16
PT-INR	1.79
APTT (s)	45.5
FDP (μg/mL)	14.2
D-dimer (μg/mL)	5.8
HIV antibody	negative

PT-INR, prothrombin time-international normalized ratio; APTT, activated partial thromboplastin time; FDP, fibrin degradation product; HIV, Human immunodeficiency virus.

lower lobe (Fig. 1). The rapid test for influenza A, sputum acid-fast stain, and urine antigen for *Streptococcus pneumoniae* and *Legionella* spp. were negative. Taking into account the results at the previous hospital, intravenous levofloxacin, vancomycin, and peramivir were started. We could not perform cerebrospinal fluid (CSF) examination because of his low platelet count.

After admission, his blood pressure decreased and his respiratory status continued to worsen, requiring intubation and transfer to the intensive care unit. Noradrenalin was administered, and blood transfusion was performed. Bone marrow examination revealed mild hemophagocytosis, without malignancy (Fig 2). Bone marrow fluid culture was negative. On day 3 of admission, both aerobic and anaerobic blood cultures yielded gram-negative rods. Brain computed tomography (CT) revealed no apparent abnormal findings (Fig. 3). Intravenous antibiotics (meropenem 6 g/day) were added because of suspected melioidosis with meningitis. However, his renal function worsened and continuous hemodiafiltration was started. On day 5, the blood culture was confirmed to contain *B. pseudomallei* via loop-mediated isothermal amplification targeting the TTS1 gene cluster, which contains sequences specific for *B. pseudomallei* [5] and via multiplex polymerase chain reaction using conserved and *B. pseudomallei*-specific primers [6]. These tests were performed at the National Institute of Infectious Diseases, Japan. *B. pseudomallei* were also detected in the sputum culture. Minimum inhibitory concentrations of antibiotics on *B. pseudomallei* are shown in Table 2. At this time, his condition had been improving, although the pancytopenia remained. Meropenem and levofloxacin were changed to ceftazidime based on the susceptibility test results. On day 10, he was extubated. We performed CSF examination with platelet transfusion, which revealed elevated protein levels (180 mg/dL), with 181.4 white blood cells/mm^3 and 67% polymorphonuclear cells, although the culture was negative. On the same day, he experienced high fever with shaking chills, and purpura appeared on his extremities. Septic emboli were suspected, and the treatment was changed to vancomycin, metronidazole, ceftazidime, levofloxacin, and liposomal amphotericin B. However, his respiratory condition and consciousness were acutely deteriorated and he required intubation again. His pupils were dilated and nonreactive. Brain CT revealed multiple occupied

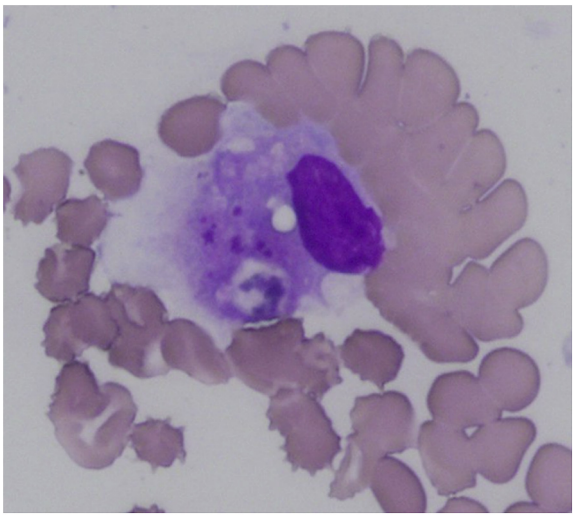

Fig. 2. Bone marrow smear showing hemophagocytosis (Giemsa stain; ×400).

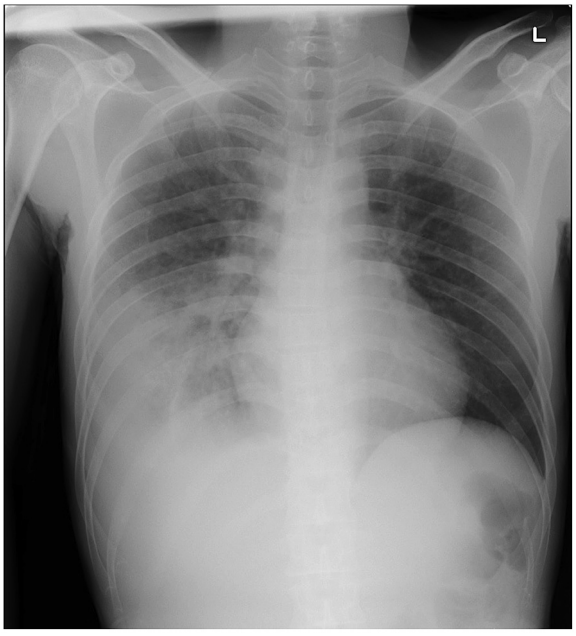

Fig. 1. Chest radiography at presentation revealed consolidation in the right lower lobe.

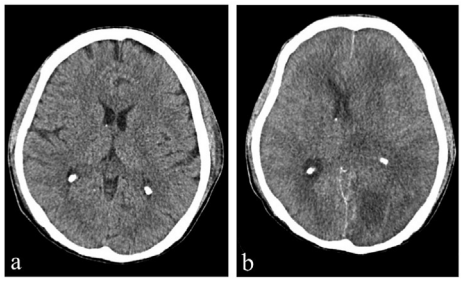

Fig. 3. (a) Initial computed tomography (CT) of the brain revealed no apparent abnormal findings. (b) Follow-up CT revealed multiple occupied lesions.