MELIOIDOSIS WITH ADRENAL GLAND ABSCESS

SAI-CHEONG LEE, TIING-SOON LING, JIH-CHANG CHEN, BI-YU HUANG, AND WEN-BIN SHEIH
Division of Infectious Diseases, Department of Emergency, and Department of Internal Medicine, Chang Gung Memorial Hospital, Keelung, Taiwan, Republic of China

Abstract. We report a case of melioidosis with left adrenal gland abscess in a 51-year-old man from Taiwan who traveled to Rangoon, Burma for a four-day tour on July 15, 1997. The patient developed fever and left upper abdominal pain upon returning to Taiwan on July 19, 1997. Ten days after returning to Taiwan, he was admitted to Chang Gung Memorial Hospital in Keelung, Taiwan and blood culture on admission was positive for Burkholderia pseudomallei. Computerized tomography of the abdomen revealed left adrenal gland swelling and suppuration. Treatment with parenteral ceftazidime and cotrimoxazole for three weeks followed by two months of oral cotrimoxazole cured the infection. The patient remained asymptomatic at 12 months follow-up.

Melioidosis is an infectious disease of animals and humans caused by Burkholderia pseudomallei and is endemic in Southeast Asia and northern Australia.1-4 The clinical presentation of melioidosis may vary from an asymptomatic infection to fulminant sepsis with organ involvement that if not appropriately treated, has a high mortality rate.5,6,7 Adrenal gland involvement is a rare complication of melioidosis and has not been well described in the literature. We describe a diabetic patient who contracted melioidosis that was complicated by adrenal gland abscess and discuss the possible route of infection.

CASE REPORT

The patient, a 51-year-old supermarket manager in Taiwan, had diabetes mellitus for three years. He had regularly taken oral hypoglycemic agents since the onset of diabetes. On July 15, 1997, he traveled to Rangoon, Burma with friends for a four-day tour involving sightseeing and golf. During the trip, he visited several temples, played golf, and went fishing in Rangoon. In accordance with local customs, he walked barefoot on entering temples. On July 19, 1997, he returned to Taiwan. On the night of his arrival in Taiwan, he developed fever and chills and took antipyretics bought from drug stores. Fever and chills persisted. He visited a local hospital hear his home four days later and was treated unsuccessfully. The next day, fullness and pain over the left upper quadrant of the abdomen developed and aggravated gradually. He consulted the emergency service at Chang Gung Memorial Hospital in Keelung, Taiwan five days later and was admitted. There was no diarrhea, dysuria, jaundice, or conscious disturbance.

Clinical examination indicated an acutely ill man. He had a temperature of 38.2°C, a respiration rate of 20/min, a pulse of 82/min, and a blood pressure of 120/70 mm of Hg. He was not anemic or icteric. His throat was normal, his neck was supple without lymphadenopathy, and his respiratory and cardiovascular functions were normal. The abdomen was soft with tenderness over the left upper quadrant and bowel sounds were normal. There was no evidence of hepatosplenomegaly or lymphadenopathy. Neurologic examination results were negative. There were no skin or muscle abnormalities.

On admission, he had a hemoglobin level of 12.9 g/dl, a hematocrit of 35.1%, a white blood cell count of 5,700/mm³ with 70.3% neutrophils, 18.9% lymphocytes, and 9.8% monocytes, and a platelet count of 153,000/mm³. Biochemical test results were as follows: blood urea nitrogen = 7mg/dl, creatinine = 1.0 mg/dl, glucose = 361 mg/dl, blood glutamic oxaloacetic transaminase = 51 units/L (normal = 0–33 units/L), glutamic pyruvic transaminase = 37 units/L (normal = 0–40 units/L), alkaline phosphatase = 111 units/L (normal = 0–100 units/L), albumin = 2.9 g/dl, and globulin = 3.0 g/dl. Serum sodium, potassium, and calcium concentrations were normal.

A chest radiograph showed an interstitial lung infiltrate (Figure 1). Urinalysis revealed only glucosuria (1.0 mg/dl). His stool was negative for occult blood or parasite ova. Blood smear was negative for malaria.

On the day of admission, typhoid fever was initially suspected and treatment with parenteral cotrimoxazole (800 mg of sulfamethoxazole and 160 mg of trimethoprim), three times a day, was initiated. The fever subsided the next day and parenteral cotrimoxazole was continued for four weeks. Continued clinical improvement was observed. Eight days after admission, blood cultures done one week before showed Burkholderia pseudomallei. A paper disk test showed that the organism was resistant in vitro to ampicillin, cotrimoxazole, and all aminoglycosides, but sensitive in vitro to chloramphenicol, ceftriaxone, cefazidime, and imipenem. Cefazidime, 1g three times a day, was added to the treatment regimen. Computerized tomography of the abdomen on the same day showed swelling and inflammation of the left adrenal gland with suppuration (Figure 2). He remained afebrile and the left upper abdominal pain improved. The blood glucose level was controlled with oral hypoglycemic agents. Follow-up computerized tomography of the abdomen two weeks after admission showed improvement of left adrenal gland swelling and suppuration. Serum cortisol levels three weeks after admission were 6.91 µg/dl at 8:00 AM and 0.87 µg/dl at 4:00 PM (normal = 4.3–22.4 µg/dl at 8:00 AM and 3.1–16.7 µg/dl at 4:00 PM). Combination therapy with parenteral cotrimoxazole and ceftazidime was given for three weeks. A repeated chest radiograph at four weeks after initiation of therapy was normal. Repeated computed tomography of the abdomen 34 days after admission showed further improvement of the left adrenal gland swelling and suppuration. He was discharged and given a regimen of oral cotrimoxazole (800 mg of sulfamethoxazole and 160 µg of trimethoprim), three times a day, on the 35th day after admission. He was followed-up regularly in our outpatient clinic. Repeated morning and afternoon serum cortisol levels were normal. The patient remained asymptomatic. Eight weeks after discharge, his follow-up computerized tomography showed further improvement of left adrenal gland swelling and suppurative cyst. The patient remained asympomatic at 12 months follow-up.
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FIGURE 1. Chest radiograph of the patient on admission, showing a bilateral interstitial infiltrate.

FIGURE 2. Computerized tomography of the abdomen of the patient on August 7, 1997, showing swelling and suppuration of the left adrenal gland. The same tomograph is shown at two different levels. Oral cotrimoxazole were given for two months. At 12-months follow-up, the patient was asymptomatic.

FIGURE 3. Abdominal computerized tomography on October 7, 1997 revealed a partial resolution of the left adrenal gland swelling (Figure 3). Oral cotrimoxazole were given for two months. At 12-months follow-up, the patient was asymptomatic.

DISCUSSION

Melioidosis was first described in 1911 in Burma and this country is now is a well-known endemic area for this disease.1,2,5 Our patient acquired melioidosis while traveling in Burma. However, the route of transmission of B. pseudomallei in this patient is not certain. Burkholderia pseudomallei is a free-living bacterium found widely in the soil and water of plant fields, drains, gardens, and playgrounds in endemic areas.1,6 This organism usually infects humans via contaminated soil or water that enters the body through a pre-existing skin abrasion or ulcer or by inhalation of infectious dust particles.1,6 This patient visited several temples in Rangoon and walked barefoot on entering the surrounding area of the temples in accordance with local habit customs. Thus, it is possible that a minor abrasion wound in his foot occurred and was not noticed while walking barefoot near the temple, and that the organism then invaded the blood stream percutaneously through such an abrasion causing acute septicemic melioidosis. However, an inhalation route is not excluded because the patient did have an interstitial lung infiltrate on admission, which may be the first radiologic evidence of pulmonary melioidosis although he had no respiratory symptom. He may have inhaled or swallowed the organism while golfing or fishing. Because B. pseudomallei is endemic throughout Southeast Asia, we recommend that travelers do not go barefoot in any developing areas. This precaution would avoid not only melioidosis, but other pathogens acquired through the skin such as hookworm and Strongyloides.

Diabetes is a recognized major pre-disposing factor for melioidosis, as in this patient.1,5,9 The clinical forms of melioidosis vary from acute septicemic form to subacute, chronic, and subclinical form.1,3,5 The incubation period can be as short as 2–5 days, as in our patient, and as long as 26 years after the initial infection.1,3,5 Melioidosis with multiple organ involvement with or without suppuration has been reported to include lung, skin and soft tissue, skeletal, hepatobiliary genitourinary, lymphatic cardiovascular, and central nervous systems, the eye, and the parotid gland.7,21 Although infection of the adrenal gland with suppuration by this organism had been mentioned in the literature, melioidosis with adrenal gland abscess had not been well described.12,13 In non-
endemic areas, the septicemic form of melioidosis without an obvious source and leukocytosis is frequently misdiagnosed. In our patient, a diagnosis of melioidosis was not made until 10 days after the onset of disease. A mortality rate of the acute disseminated septicemic form of melioidosis as high as 87% has been reported for patients who do not receive appropriate treatment. Since foreign travel is becoming increasingly common in developing and developed countries, general practitioners should be aware of the diagnosis and treatment of melioidosis.

According to the literature, B. pseudomallei is sensitive to chloramphenicol, cotrimoxazole (trimethoprim-sulfamethoxazole), ceftazidime, and imipenem. In this patient, B. pseudomallei isolated from blood was sensitive to all of these antimicrobial agents except for cotrimoxazole. However, cotrimoxazole appeared to be effective clinically in this case and was therefore continued until the completion of treatment. The results of in vitro susceptibility tests results sometimes do not correlate with clinical efficacy. Ceftazidime was added to the regimen of parenteral cotrimoxazole and was given for three weeks in the acute septicemic stage, followed by oral cotrimoxazole for two months in the non-septicemic stage. This is the minimum duration of treatment of melioidosis recommended in the literature. It is still possible that ceftazidime given for three weeks was the curative agent and cotrimoxazole was mainly bacteriostatic. We shall follow-up this case for several years to determine whether the two-month course with oral cotrimoxazole was adequate.

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Authors’ addresses: Sai-Cheong Lee, Division of Infectious Diseases, Chang Gung Memorial Hospital, 222 Mai Chin Road, Keelung, Taiwan, Republic of China. Tieng-Soon Ling and Jih-Chang Chen, Department of Emergency, Chang Gung Memorial Hospital, Keelung, Taiwan, Republic of China. Bi-Yu Huang and Wen-Bin Sheih, Department of Internal Medicine, Chang Gung Memorial Hospital, Keelung, Taiwan, Republic of China.

Reprint requests: Sai-Cheong Lee, Division of Infectious Diseases, Chang Gung Memorial Hospital, 222 Mai Chin Road, Keelung, Taiwan, Republic of China.

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