Case Report: Non-septicemic Melioidosis Presenting as Cardiac Tamponade

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Abstract. Melioidosis is endemic in Taiwan. It is caused by infection with Burkholderia pseudomallei. A prolonged course of oral eradication therapy to avoid relapse after an intensive intravenous therapy is recommended to treat melioidosis. Melioidosis with cardiac involvement is rare and is often combined with septicemia, for which the mortality rate is 20–60%. The initial clinical presentations of melioidosis mimic Mycobacterium tuberculosis infection, which is the most common etiology of bacterial pericarditis in Taiwan. We present a case of non-septicemic melioidosis that presented as non-suppurative cardiac tamponade and left subcarinal lymphadenopathy. Underlying diseases included hepatitis B–related liver cirrhosis and hepatocellular carcinoma. The patient was successfully treated with 2 weeks of intravenous ceftazidime and 12 weeks of oral doxycycline, trimethoprim-sulfamethoxazole, and amoxicillin/clavulanate. Melioidosis-related pericarditis should be considered in the differential diagnoses of bacterial pericarditis in Taiwan.

INTRODUCTION

Melioidosis, a disease caused by Burkholderia pseudomallei infection, is endemic in Taiwan. Most patients present with pneumonia, bacteremia, genital-invasive infection, and skin and soft tissue infection. Although isolated pericarditis had been reported in one diabetic patient, most patients presented with cardiac tissue involvement combined with septicemia. The mortality rate of septicemic melioidosis is 20–60%. Diabetes mellitus, pre-existing renal disease, thalassemia, and occupational exposure are the most common risk factors for acquiring melioidosis. An immunocompromised status of the host is also a risk factor. We present a rare case of B. pseudomallei infection with initial clinical presentation of non-suppurative cardiac tamponade and left subcarinal lymphadenopathy who had underlying hepatitis B–related liver cirrhosis and hepatocellular carcinoma.

CASE REPORT

The patient was a 73 year-old man, a chronic hepatitis B carrier, and a retired official of an agricultural cooperative for 10 years. He traveled to Mainland China 20 years earlier and has been well in recent years. Thirty days before admission, he suffered from an episode of fever, which was followed by a loss of body weight and a persistent yellowish productive cough. He presented to our chest clinic 2 weeks before hospitalization. The chest radiography and the chest computed tomography (CT) scan showed one calcified nodule in the left upper lung (Figure 1A) and two lymph nodes with central necrosis below the left subcarinal area (Figure 1B). The results of staining for sputum acid fast bacilli were negative. Abdominal ultrasonography showed liver cirrhosis with multiple hepatic tumors. The level of α-fetoprotein was 5,208 ng/mL (reference range, 0–13.4 ng/mL). The patient was admitted with the diagnosis of hepatocellular carcinoma (HCC) with lung metastasis. On admission, his ear temperature was 38.8°C, respiratory rate was 20/min, and pulse rate was 96/min. His blood pressure was 120/74 mm of Hg. On physical examination, his jugular veins were engorged, and his heart was enlarged with an irregular rhythm and a loud pericardial rub. The chest radiograph on the date of admission showed a rapid enlargement of a globular heart shadow. The abdominal CT showed massive pericardial effusion (Figure 1C) and three arterial blushed enhancing nodules in the liver that were compatible with HCC. An echocardiogram showed a large amount of pericardial effusion with right ventricular collapse, and cardiac tamponade was our impression. As an emergent maneuver, 250 mL of non-coagulable, turbid, reddish pericardial effusion was drained. The white blood cell (WBC) count in the pericardial effusion was 800/mm³ (neutrophil/lymphocyte/monocyte ratio: 50/38/12). The pericardial effusion to serum ratio of total protein and lactate dehydrogenase (LDH) was, respectively, 0.76 (5.6/7.4) and 4.55 (2,355/518). Gram and acid fast bacilli staining, bacterial and Mycobacterium tuberculosis (TB) cultures, and tests for malignancy cells in the pericardial effusion were all negative. Two sets of cultures for bacteria in the blood were negative. HCC invasion of the pericardium complicated by acute pericardial effusion accumulation was suspected. Although this patient had a fever on the admission date, an infectious disease was not favored initially because the fever subsided spontaneously without antibiotic treatment, and no other clinical signs of infection were seen. He received transarterial chemoembolization therapy for HCC on the eighth hospital day. Unexpectedly, the pericardial effusion culture indicated the presence of a strain of B. pseudomallei that was susceptible to amoxicillin/clavulanate, ceftazidime, imipenem, and trimethoprim-sulfamethoxazole (TMP-SMX). He received intravenous ceftazidime 2 g every 8 hours for 14 days and then received 12 weeks of a combination of doxycycline 100 mg twice a day, TMP-SMX (80–400 mg) three tablets twice a day, and amoxicillin/clavulanate (250/125 mg) two tablets every 8 hours. The follow-up chest radiography and chest CT scan after complete therapy showed a normal heart size without left hilar lymphadenopathy. There was no evidence of relapse of the melioidosis at 1-year follow-up.

DISCUSSION

The most frequent extrahepatic metastases of HCC are lung, lymph nodes, and adrenal glands. Cardiac tamponade caused by spontaneous rupture of a mediastinal lymph node...
metastasis of HCC has also been reported. In our patient, a recent afebrile condition with relative low WBC counts and normal neutrophil to lymphocyte ratio in the pericardial effusion resulted in an initial misdiagnosis of HCC with metastasis to left upper lung, mediastinal lymph nodes, and pericardium metastasis.

Elevated LDH and total protein levels of the pericardial effusion in this patient hinted at bacterial pericarditis. Staphylococcus spp., Streptococcus spp., Haemophilus spp., and M. tuberculosis are the most frequent pathogens of bacterial pericarditis. Tuberculosis is the leading infectious cause of death in Taiwan, and M. tuberculosis is the most likely pathogen in constrictive pericarditis in Taiwan. The portal of entrance of tuberculosis might be the mediastinal lymph nodes. The initial clinical presentations of melioidosis can mimic tuberculosis, which exhibits fever, dyspnea, a loss of body weight, and fatigue. The clinical course of melioidosis and tuberculosis includes acute fulminant stage and chronic subclinical infection. It might be difficult to distinguish one from the other. Accordingly, a misdiagnosis of tuberculosis in a melioidosis patient can easily occur in Taiwan.

The incubation period of melioidosis is 1–21 days, but it can be > 60 years. Multiple risk factors, including diabetes mellitus, pre-existing renal disease, thalassemia, and occupational exposure, have been confirmed for melioidosis. Hilar lymphadenopathy would be a rare form of chronic infection and a re-activation of melioidosis, which might be associated with a decrease in host immune status, such as liver cirrhosis. Occupational exposures in Taiwan, a travel history to mainland China, and an immunocompromised condition of liver cirrhosis and HCC were the major risk factors in this patient.

A 14-day course of ceftazidime alone was used in our patient because addition of TMP-SMX to ceftazidime during initial intensive therapy of severe melioidosis does not reduce the acute mortality rate. The disease relapse rate was ~13%, and the mean duration before relapse was 8 months if a non-standard regimen was prescribed. The current recommendation therapy in eradication phase is a three-drug regimen (TMP-SMX and doxycycline) for 20 weeks which is as effective as and better tolerated than a four-drug regimen (TMP-SMX, doxycycline, and chloramphenicol). Because the three-drug regimen was just recently recommended, we were uncertain about its efficacy against invasive melioidosis infections, such as pericarditis, and we elected to use a four-drug regimen. There was a shortage of chloramphenicol in our hospital, and thus amoxicillin/clavulanate was used instead of chloramphenicol. This patient complained of dizziness after oral combination therapy, and a 12-week course rather than a 20-week course was prescribed. A rationale for combining the three-drug regimen with amoxicillin/clavulanate was unclear, and this combination may cause the unbearable side effect that led to the early cessation of the eradication therapy in our patient.

In conclusion, melioidosis pericarditis can be misdiagnosed as M. tuberculosis pericarditis in areas, such as Taiwan, where both diseases are endemic. Non-septicemic cardiac tamponade with subcarinal lymphadenopathy is a rare form of melioidosis-associated pericarditis. A differential diagnosis of melioidosis should be considered for pericarditis patients in Taiwan. Although a 20-week course of oral eradication therapy is recommended after 2 weeks of intravenous intensive therapy, our melioidosis pericarditis patient was successfully treated with a 12-week course.

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