Case Report

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비장 경색을 동반한 브루셀라증 1례

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Splenic Infarction Associated with Brucellosis in a Non-Endemic Area

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A 45-year-old man was referred from a local clinic with persistent fever, intermittent pain in the left upper abdomen, and weight loss of 7 kg. He quit his animal husbandry 18 months ago when his cows were found to be infected with *Brucella*. Abdominal computed tomography (CT) scan taken on admission showed splenomegaly with a wedge-shaped hypoattenuating region in the enhanced image, which was consistent with splenic infarction. Serology for *Bruculla* was strongly positive with the standard tube agglutination test (1/2560). After initiation of doxycycline (100 mg every 12 hrs) and rifampin (600 mg every day), the patient's condition improved, and was discharged with oral antibiotics that were to be continued for 3 months. During the 12 months' follow up at the outpatient department, the patient had no symptoms, and the last agglutination titer for *Brucella* in serum had decreased to 1/40. To our knowledge, this is the first report on splenic infarction associated with brucellosis in Korea, which was treated successfully with antibiotic therapy.

Key Words: Brucellosis, splenic infarction

Introduction

Human brucellosis has a wide spectrum of clinical manifestations and is characterized by various non-specific symptoms. Brucellosis can involve almost any organ in the body, but involvement of the vascular system is quite rare (1). Herein, we present a case of splenic infarction in a patient with brucellosis with a review of the literature.

Case Report

A 45-year-old male was referred to our hospital for evaluation of fever, chills, fatigue, headache, arthralgias in both knees, intermittent pain in the left upper

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abdomen, and weight loss of 7 kg during the past 2 months. He had been admitted to a local clinic due to the aforementioned symptoms and was given broad spectrum antibiotics for 7 days, but his symptoms did not improve. His past history was positive for diabetes, which was diagnosed 8 years ago and managed with oral hypoglycemic agents. He stopped animal husbandry 18 months ago when he found out that his cows were infected with *Brucella*; serologic tests performed at that time for brucellosis were negative. On admission to our hospital, the patient appeared chronically ill, but his vital signs were stable, and the body temperature was 37.3℃. His heart sound was normal. Abdominal examination revealed tenderness in the left hypochondrium. There were no skin rashes or lymphadenopathy. Laboratory test results were as follows: hemoglobin, 11.8 g/dL; white blood cell count, 3,230/mm^3; platelet count 24,000/mm^3; protein, 8.4 g/dL; albumin, 3.6 g/dL; bilirubin, 0.73 mg/dL; alanine transaminase, 47 IU/L; aspartate transaminase, 108 IU/L; alkaline phosphatase, 1,284 IU/L; BUN, 9.0 mg/dL; creatinine, 1.0 mg/dL; lactose dehydrogenase, 807 IU/L; erythrocyte sedimentation rate, 50 mm/h; and CRP, 27.9 mg/dL. Simple chest x-ray showed no active lung lesions but abdominal computed tomography (CT) scan showed splenomegaly with a wedge-shaped hypoattenuating region in the enhanced image (Fig. 1), which was consistent with a splenic infarction. Echocardiography showed no abnormalities of the valves. Serology for Brucella was strongly positive with the standard tube agglutination test (1/2,560), but nothing grew from the blood cultures taken on admission. The protein C, protein S, and antithrombin Ⅲ levels were normal, and antiphospholipid antibodies and lupus anticoagulant were not detected. We examined the bone marrows to exclude other causes of pancytopenia, and the bone marrow examination was consistent with a reactive marrow.

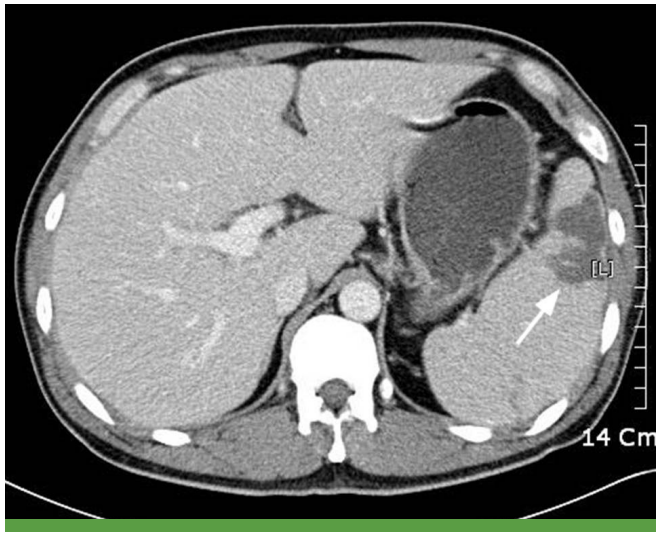

Figure 1. Post-contrast CT showing low-attenuation lesions (arrow) in the spleen.

Treatment was initiated with doxycycline (100 mg every 12 hrs) and rifampin (600 mg every day). After 3 days of treatment, his symptoms began to improve. The patient was discharged on the 11th hospital day and was scheduled to continue treatment for 3 months with oral antibiotics. After two months, the agglutination titer for Brucella in serum had dropped to 1/640 and hematologic abnormalities, such as reversed A/G ratio, returned to normal. Complete blood cell count and the liver function test results also normalized. On the follow-up CT scan, the spleen was found to have markedly decreased in size with the infarcted area having turned fibrotic. During the 12 months' follow up, the patient was free of symptoms, and the last agglutination titer for Brucella in serum was found to have decreased to 1/40.

Discussion

Brucellosis is a zoonosis, occurring predominantly in the Mediterranean basin, Latin America, Africa, the Middle East, and South Asia, but it rarely occurs in the East Asian countries. The rarity of cases and the nonspecific clinical presentation of brucellosis may result in delayed diagnosis in East Asia. Although *Brucella* melitensis is the most frequent species in endemic countries (2), *Brucella* abortus is the major pathogenic species in Korea (3).

Cardiovascular involvement of brucellosis is rare, occurring in only 1% to 2% of the cases (1). Of the complications related to the cardiovascular system, endocarditis is the main cause of morbidity and mortality related to brucellosis. In Korea, 3 cases of Brucella endocarditis have been reported, and two of these cases developed metastatic infections, such as splenic abscess and spondylitis (4-6). In our patient solitary splenic infarction occurred without evidences of endocarditis. Associated vascular complications, such as arterial aneurysm formation (7), and thrombosis of the veins or arteries (8), are extremely rare. Moreover, splenic infarction by brucellosis has only been reported in the English literature (9).

Suggested mechanisms for vascular involvement include septic embolism, endophlebitis, induction of inflammation, damage to the adjacent tissue during an infectious process, direct endothelial damage, and induction of a transient hypercoagulable state (8, 9). In our patient, studies for a hypercoagulable state were negative and the cardiac investigation results were also normal. Therefore, direct endothelial damage seems to be the most plausible explanation in the present case.

Splenic infarction generally presents with nonspecific symptoms

such as abdominal pain, nausea, and bloating (9); our patient experienced intermittent pain in the left upper abdomen.

The diagnosis of human brucellosis is based on isolating Brucella by blood culture (10) and on demonstrating specific antibodies through serologic tests (11). In our patient, the serology for *Brucella* was strongly positive, but the blood and bone marrow cultures were negative, which may have been the result of previous antimicrobial exposure to netilmicin.

The diagnosis of splenic infarction is based on imaging studies (12). CT is the imaging modality of choice (13), and visualization of a low density lesion without uptake of intravenous contrast is highly suggestive of splenic infarction.

The management of splenic infarction should be conservative, reserving surgery for complicated case (9). Although some authors have reported favorable results with anticoagulant, especially in the presence of antiphospholipid antibodies, the benefit of its administration is still controversial (14).

Doxycycline administered for 6 weeks with streptomycin for 2–3 weeks, or rifampicin for 6 weeks, is the standard therapy for brucellosis (15). If the patient shows endocarditis, spondylitis, or neurobrucellosis, prolonged therapy of 6 to 52 weeks is required (15). In a previously reported case of splenic infarction, the patient was given a prolonged course of antibiotics for 12 weeks (9).

In human brucellosis, splenic infarction can developed as one of the vascular complications, and therefore, brucellosis should be included in the differential diagnosis of splenic infarction.

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