

Spontaneous Splenic Rupture with Hematoma in A Patient with Brucellosis

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Brucellosis is one of the most common zoonotic diseases worldwide with a variable frequency in European countries. Brucellosis is usually transmitted after direct contact with or consumption of dairy products from an infected animal. Initially, our patient, a 65 year-old man, had nonspecific manifestations of fever, malaise, headache, anorexia and arthralgia which are the classic symptoms of this disease. After the diagnosis of brucellosis had been confirmed by blood culture and serology, progressive thrombocytopenia developed in spite of appropriate antibiotic administration. Radiological investigation revealed imaging findings consistent with splenic rupture. The complete recovery was observed after administration of antibiotic therapy and platelet transfusion. Spontaneous rupture of the spleen with subcapsular hematoma is an extremely rare condition in brucellosis. We want to remind clinicians of this rare complication. (*Chang Gung Med J 2011;34(6 Suppl):52-5*)

Key words: brucellosis, splenic rupture

Human brucellosis is a potentially life-threatening multisystemic disease. The global burden of human brucellosis remains enormous; it causes more than 500,000 infections per year worldwide.⁽¹⁾ Brucellosis remains the most common zoonotic infection worldwide, including Turkey. Brucellosis in humans is usually associated with occupational exposure to infected animals, such as by laboratory workers, veterinarians and slaughterhouse workers, or the consumption of unpasteurized dairy products. Although brucellosis in humans is rarely fatal, it may have severe complications.⁽²⁾ In this report, we present a rare case of spontaneous splenic rupture caused by brucellosis.

CASE REPORT

A 65-year-old man was admitted to the infectious disease ward because of persistent fever, chills,

sweating, malaise, anorexia, weight loss, nausea, vomiting, and headache for five days. He did not smoke or drink and had no history of drug abuse. There was no history of travel or exposure to ill persons. He had been well and did not take any medication before this hospitalization. He denied any recent history of animal contact, but he had eaten raw milk cheese before this episode.

On admission, he had a temperature of 39.8°C, blood pressure of 110/70 mm Hg, pulse rate of 125 beats/min, and respiratory rate of 24 breaths/min. He had no skin or mucosal lesions. On physical examination, dullness to percussion of Traube's space was detected. The liver was palpated approximately 2 cm below the right costal margin. The remainder of the examination had no remarkable findings. His hematocrit was 39.2%, white blood cell count was $7.4 \times 10^3 / \mu\text{L}$ with 64% neutrophils, and platelet (PLT) count was $86 \times 10^3 / \mu\text{L}$ (normal range, 150-450 \times

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$10^3 / \mu\text{L}$). The erythrocyte sedimentation rate was 10 mm in 1 hour. Liver function tests were abnormal as follows: aspartate aminotransferase 266 U/L (normal range, 7–42 U/L), alanine aminotransferase 281 U/L (normal range, 2–54 U/L), gamma-glutamyl-transpeptidase 241 U/L (normal range, 11–50 U/L), and alkaline phosphatase (ALP) 365 U/L (normal range, 91–258 U/L). All other laboratory findings were within normal limits. The chest radiograph was normal. Screening and confirmative serological tests for brucellosis were done because of his history of raw milk cheese consumption. A Brucella Rose Bengal agglutination slide test (Seromed Laboratory Products, Istanbul, Turkey) was positive, and a Brucella Standard Agglutination Test (Pendik Veteriner Kontrol Enstitüsü, Istanbul, Turkey) was reactive with a titer of 1:640. Two sets of blood cultures grew bacteria (BACTEC 9050, Becton Dickinson, Franklin Lakes, NJ, U.S.A.) following 72 h of incubation at 37°C. Gram staining of the bacterial colonies on 5% sheep blood agar revealed small gram-negative coccobacilli. The organism was confirmed to be *B. melitensis* by standard biochemical reaction and API 20 NE (Biomerieux, Marcy-J'Etoile France). Treatment was initiated on the second day of hospitalization, with a combination of 200 mg doxycycline plus 600 mg rifampin every day for 6 weeks. Four days after antibiotic treatment started, his body temperature was 38°C and epistaxis developed. Thrombocytopenia was found with a PLT

count of $15 \times 10^3 / \mu\text{L}$. Therefore, the patient was given two units of platelet suspension. The next day, the PLT count was $20 \times 10^3 / \mu\text{L}$.

An ultrasound of the abdomen demonstrated modest liver and spleen enlargement in addition to free intraperitoneal fluid. Subsequent contrast-enhanced abdominal computed tomography (CT) of the abdomen revealed an ill-defined heterogeneous subcapsular heterogeneous parenchymal lesion adjacent to focal regional capsular irregularity in the spleen, along with hepatosplenomegaly and free fluid (Fig. 1). Abdominal magnetic resonance imaging (MRI) obtained after CT scanning demonstrated that the splenic lesion was hemorrhagic and there was also a focal capsular defect next to the lesion (Fig. 2). As the patient denied a history of recent abdominal trauma, it was concluded that imaging findings showed splenic interstitial hemorrhage and rupture secondary to brucellosis.

Other possible causes of spontaneous rupture of the spleen in addition to brucellosis were investigated. A heterophile antibody test, Epstein Barr viral capsid antigen (EBVCA) IgM, EBVCA IgG, and cytomegalovirus (CMV) IgM and IgG were negative. The splenic rupture was considered to be a complication of brucellosis. The regimen of doxycycline plus rifampin was continued. The PLT level was kept above $30 \times 10^3 / \mu\text{L}$ using PLT transfusion only when needed. A total of four units of PLTs were transfused. On the 10th day of treatment, his body tem-

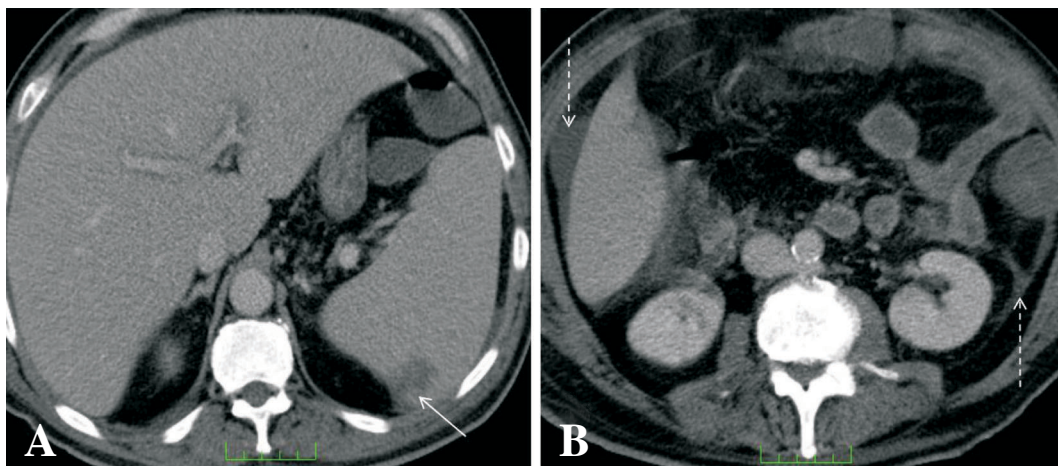


Fig. 1 Contrast-enhanced abdominal computed tomography, transverse images: There is hepatosplenomegaly and an ill-defined heterogeneous hypodense lesion in the spleen with focal irregularity of the adjacent capsule (A, arrow). Also note the presence of free intraperitoneal fluid (consistent with hemorrhage) (B, arrows).

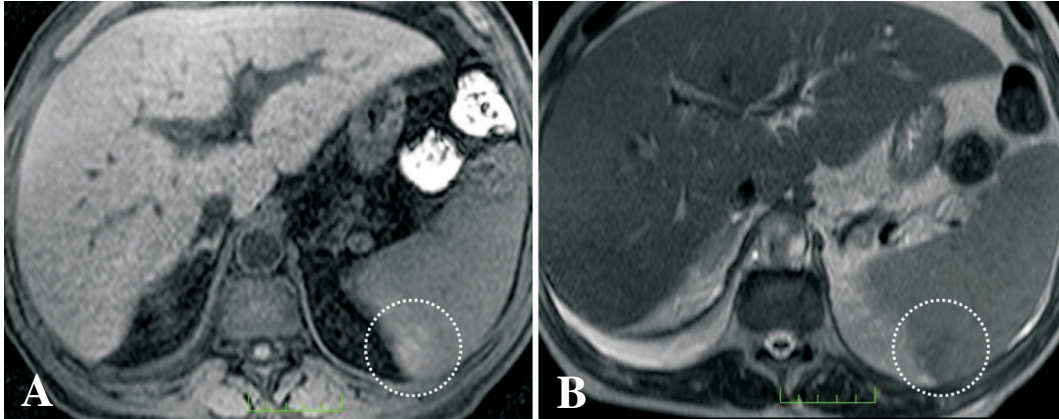


Fig. 2 Abdominal magnetic resonance imaging, transverse images. The splenic lesion (in circle) is hyperintense on T1W images (A) and hypointense on T2W images (B), consistent with hemorrhage.

perature was 36.4°C; the PLT count was $134 \times 10^3/\mu\text{L}$. Combination therapy was continued for six weeks and the patient recovered completely.

DISCUSSION

Brucellosis is the most common zoonotic infection worldwide. This disease is usually benign, but complications may lead to an increased risk of mortality and morbidity. Spontaneous splenic rupture secondary to brucellosis is an extremely rare but important clinical condition, with only three cases reported in the literature.⁽³⁻⁵⁾ To the best of our knowledge, there is no report based on imaging studies in the English literature on the diagnosis of splenic rupture and free intraperitoneal fluid consistent with hemorrhage secondary to brucellosis. Atraumatic rupture of the spleen has been described in the medical literature as a clinical situation which may carry an increased risk of morbidity and mortality if unrecognized and untreated. Some authors have suggested that atraumatic or so-called spontaneous rupture of a diseased spleen should be termed “pathologic rupture”. The incidence of spontaneous rupture is unknown.⁽⁶⁾

Spontaneous splenic rupture is rare, but often life-threatening. There is no guidance for its management. Many infectious agents are thought to play an important role in the process of spontaneous splenic rupture. Infectious mononucleosis, CMV infection, human immunodeficiency virus infection, tuberculosis, malaria, dengue fever, *Babesia microti* infection,

and *Bacillus* species infection have been associated with spontaneous splenic rupture.⁽⁷⁻¹³⁾ The most common symptoms of spontaneous splenic rupture are fever, general malaise, chills, left upper quadrant pain, and nausea. Physical examination of the abdomen reveals tenderness and often a palpable tender mass in the left upper quadrant.^(7,9,10,12) A meta-analytic review revealed that infection was one of the six major etiologies of spontaneous splenic rupture and 6.6% of patients had bacterial infection.⁽⁷⁾ Our case is the fourth case of spontaneous splenic rupture associated with brucellosis in the literature. Different mechanisms may contribute to spontaneous rupture of the spleen, such as splenomegaly due to pathological infiltration, especially, of the capsule; splenic infarct with subcapsular hemorrhage and subsequent rupture; and coagulopathy. However, rupture is usually the result of a combination of mechanisms.⁽⁸⁾ The splenic capsule is thin and fragile. During infectious processes, there may be congestion and dilatation in the sinusoids and Billroth cords as well as capillary thrombosis and focal necrosis in the splenic pulps. It has been suggested that because of these changes, the spleen becomes more susceptible to hemorrhage and rupture. We assume that similar mechanisms may have been responsible in our case.

Fever, nausea, vomiting and abdominal pain are the most frequent findings in spontaneous splenic rupture.^(6,8,9) In our case, all symptoms except for abdominal pain were present. Although there was no severe abdominal pain, blood count abnormalities were consistent with splenic rupture in brucellosis.

Because of the rarity of spontaneous splenic rupture, no clear consensus of treatment has been reached. In the setting of hemodynamic instability, a splenectomy is the procedure of choice. However, the choice of treatment in stable patients is controversial. Many successful cases of conservative treatment have been reported in the literature. A review of these cases, showed that conservative therapy failed in patients with persistent hemodynamic instability requiring transfusion of more than 4 units of packed red blood cells.⁽¹³⁾ In our case, the hemoglobin level was 9 g/dl, and on follow-up, it did not fall below that level. Because of significantly low levels of PLT, four units of PLTs were transfused. Our patient recovered completely with medical therapy (doxycycline plus rifampin) and PLT transfusions.

Although splenic rupture is an extremely rare complication of brucellosis, it should be kept in mind in the process of the disease, especially in patients with unexpected thrombocytopenia. Abdominal CT scanning is the imaging modality of choice in the diagnosis of this complication, while MRI may provide complimentary information. Conservative treatment should be considered initially.

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