Hemophagocytosis and pulmonary involvement in brucellosis

We report the case of a 45-year-old woman who presented with fever, hepatosplenomegaly, jaundice, rash, and tachypnea. Wright’s serum agglutination and 2-mercaptoethanol (2-ME) tests were both positive in the range of 1/2560 and 1/160 in a titer of 1/160 in. A positive Wright’s agglutination test in a titer of 1/160 in the blood sample was also noted. Four days after admission laboratory tests revealed pancytopenia: platelet count 12 × 10^9 cells/L, Hb 8.5 g/dL, and WBC 3.5 × 10^9 cells/L. Bone marrow aspiration revealed an increase in histiocytic lineage and phagocytosis of nucleated red blood cells and platelets. The cultures of bone marrow were negative. Other laboratory tests revealed negative blood cultures and serum ferritin > 1000 ng/mL.

Five days after admission, high resolution computed tomography of the chest was performed due to persistent tachypnea and rales, and showed consolidation in both lungs. After three days of treatment with hydrocortisone (200 mg/day) and anti-brucellar drugs, the platelet count increased, liver enzymes decreased (SGPT 130 IU/L, SGOT 91 IU/L), and prothrombin time (PT) and partial thromboplastin time (PTT) returned to normal.

Mild hematologic abnormalities are common in brucellosis and usually subside with treatment of the disease itself. Thrombocytopenia may be severe and associated with purpura and bleeding. Steroids have been recommended in severe thrombocytopenia associated with human brucellosis.1 Severe disorders such as hemophagocytic syndromes have also been described.2 Hemophagocytic syndrome is associated with a broad spectrum of diseases such as viral, bacterial and mycobacterial diseases.3 Signs and symptoms of hemophagocytic syndromes are often similar to common infections or mimic fever of unknown origin or hepatitis. Fever, splenomegaly, and hepatomegaly are the most common clinical findings and lymphadenopathy, jaundice, and rashes can also be found. Hemophagocytic syndrome may result in pulmonary involvement.4

In daily practice, the diagnosis of brucellosis is established by a positive Wright’s agglutination test in a titer of > 1/160 in association with an appropriate clinical setting.5 The brucella infection in our patient was established with strongly positive serology (standard tube agglutination (STA) test).
Agglutination in the presence of 2-ME was used to distinguish specific IgG (suggestive of active disease) and IgM reactivity. The test was performed in the same manner as the STA test except that sera were previously exposed to 2-ME.

The patient presented with hemophagocytic syndrome associated with brucellosis, which progressively recovered with anti-brucellar drugs and a three-day administration of hydrocortisone, leading to improvement of all hematologic, hepatic, and pulmonary changes. The large ecchymosis associated with thrombocytopenia, pulmonary involvement associated with brucella or hemophagocytosis (with negative chest X-ray), and rapid response to the short course of steroids as well as the anti-brucellar drugs were unique features of our patient. It should be said that in endemic areas, rare complications of common diseases should always be taken into account. Also, in patients with brucellosis, respiratory findings such as unexplained tachypnea accompanying a normal chest X-ray is an alarming sign pointing to a diagnosis of pneumonia, hemophagocytic syndrome, or pulmonary thromboemboli, and performing complementary radiographic imaging such as a chest CT scan may be useful and revealing.

Conflict of interest: No conflict of interest to declare.

References


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