Recurrent Epistaxis and Bleeding as the Initial Manifestation of Brucellosis

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Abstract- Severe thrombocytopenia with bleeding is rarely reported in children with brucellosis, and recurrent epistaxis is extremely rare. Brucellosis with hemorrhage should be differentiated from viral hemorrhagic fever, malignancy, and other blood disorders. Bone marrow aspiration (BMA) is mandatory to differentiate from other blood diseases. An 8-year-old boy was admitted with recurrent epistaxis, petechiae and purpura on face and extremities and bleeding from the gums. During the hospitalization, he was febrile and complained of muscle pain. Leukopenias associated with thrombocytopenia were observed. BMA showed to be normal. Among the multiple tests requested, only serum agglutination test (SAT) and 2-MercaptoEthanol test (2-ME) were positive. He was treated with Intravenous immunoglobulin (IVIG) associated with co-trimoxazole and rifampin. Finally, fever subsided, and he was discharged with good condition and normal platelet count. Brucellosis should be a differential diagnosis in patients with fever and bleeding disorders and a history of consumption of unpasteurized dairy, in endemic areas.

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Introduction

Brucellosis is a public health problem. The majority of cases of brucellosis in children occur following the consumption of unpasteurized dairy products (1). One of the common presentations of this illness is a fever of unknown origin (FUO) and the classic triad of fever, arthralgia/ arthritis, and hepatosplenomegaly that most patients show (1).

Hematologic abnormalities such as pancytopenia are common in the course of brucellosis. Sometimes, thrombocytopenia can be severe, resulting in hemorrhage into the skin and from mucosal sites (2). Various serologic tests have been used for the diagnosis of brucellosis. The serum agglutination test (SAT) is the most widely used and 2-MercaptoEthanol test (2-ME) is used to confirm the diagnosis. It is important that all titers should be interpreted on the basis of history and physical examination (1-3).

Oral doxycycline is the drug of choice and should be administered with rifampin but for children younger than eight years trimethoprim-sulfamethoxazole instead of doxycycline is recommended (3). Pasteurization of dairy products is recommended to prevent the disease, especially in children (1-3).

Case Report

An 8-year-old boy was admitted with recurrent epistaxis, petechiae and purpura on face and extremities, and gingival bleeding at the pediatric ward of Ayatollah Mousavi Hospital, Zanjan on January 2014. During the hospitalization, he was febrile and complained of muscle pain and weakness. At physical examination, he did not have any significant lymphadenopathies or organomegaly. Abdominal ultrasound was normal. Leukopenia (2.1×103/mm) and thrombocytopenia (7×103/mm) were detected in the para-clinic evaluation. BMA was done to rule out malignancies or other bone marrow disorders. The bone marrow aspiration was normal. The consultation was requested with the Pediatric Infectious Disease Specialist due to the unknown cause of fever and normal bone marrow aspiration. He recommended a brucellosis work-up, due to the history of consumption of unpasteurized dairy, persistent fever, chills and muscle pain. The results of Wright test (SAT) and 2-ME test were positive for brucellosis infection (respectively 1/640 and 1/320). The child was treated with IVIG (400mg/kg/day) for 5 days and anti-brucellosis drugs (include rifampin with a dose

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of 20 mg/kg/day and co-trimoxazole with a dose of 10 mg/kg/day based on trimethoprim). The patient's fever subsided on the sixth day of treatment. Platelet count increased up to 71×103/mm on the 6th day of treatment and up to 187×103/mm on the 10th day of treatment. The patient was discharged in good condition and was recommended continuing the anti brucellosis drugs for up to six weeks. Results of Laboratory tests were summarized in Table 1.

Table 1. Results of Laboratory tests

Tests	Patient	Normal Range
WBC	2.1×10^9/L P = 48%, L = 48%	$4.0\text{-}10.0\times10^9$/L (P = 20-45%, L = 42-77%)
HB	10.1 gr/dl	12-14 gr/dl
PLT	7×10^9/L	$150\text{-}450\times10^9$/L
ESR	55 mm/h	10 -15 mm/h
Retic	1.7%	1-1.5%
Direct Coombs	Negative	Negative
In Direct Coombs	Negative	Negative
PT	13 Sce	12-15 Sec
INR	1	1.0
PTT	35 Sec	24-40 Sec
SAT	1/640	Negative
2-ME test	1/320	Negative

WBC, White Blood Cells; Hb, Hemoglobin; PLT, Platelet Count; PT, Prothrombin Time; PTT, Partial Thromboplastin Time; Retic, Reticulocyte Counts; ESR, Erythrocyte Sedimentation Rate; SAT, Serum Agglutination Test; 2-ME, 2-MercaptoEthanol test

Discussion

The causes of severe thrombocytopenia in brucellosis are various, including hypersplenism, disseminated intravascular coagulation (DIC), bone marrow suppression, hemophagocytosis, and immune mechanisms (2). For diagnosis of this rare presentation of brucellosis such as severe thrombocytopenia and mucosal bleeding, a strong clinical suspicion is necessary. This condition should be differentiated from viral hemorrhagic fever, malignancy, and other blood diseases. Brucellosis should be a differential diagnosis in patients with fever and bleeding disorders and a history of consumption of unpasteurized dairy, especially in endemic areas. Brucellosis has a specific diagnostic test. Therefore early detection and treatment of disease is possible. Because immune mechanisms may co-exist with the disease, treatment with IVIG can be administrated with anti-brucellosis drugs. Our patient was admitted because of recurrent epistaxis which is not mentioned in previous articles (4-6). Unlike our case, the majority of previous cases had organomegaly (4-6).

Most cases of severe thrombocytopenia associated with brucellosis respond well to the antibiotic treatment with or without IVIG and recover a few days after treatment (4-6). Therefore, accurate diagnosis and prompt treatment are essential because early treatment is associated with a rapid recovery.

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