Published online 2016 January 2.

**Case Report** 

# Gum and Nose Bleeding as a Presentation of Pediatric Brucellosis

## Ali Hosseininasab<sup>1,\*</sup>; Zahra Farahmandinia<sup>2</sup>; Mahdieh Eslamian<sup>2</sup>; Pegah Arfa<sup>2</sup>; Motahareh Abbaszadeh<sup>2</sup>

<sup>1</sup>Infectious and Tropical Diseases Research Centre, Kerman University of Medical Sciences, Kerman, IR Iran

<sup>2</sup>Kerman University of Medical Sciences, Kerman, IR Iran

\*Corresponding author: Ali Hosseininasab, Infectious and Tropical Diseases Research Centre, Kerman University of Medical Sciences, Kerman, IR Iran. Tel: +98-342322250, Fax: +98-3413222763, E-mail: ali4221@kmu.ac.ir

Received: June 14, 2014; Revised: July 30, 2014; Accepted: September 5, 2014

Introduction: Brucellosis symptoms are nonspecific; the most common complaints include fever, sweats, anorexia, headache, malaise, and arthralgia. Hematological manifestations of active brucellosis vary from mild anemia and leukopenia to thrombocytopenia and rarely pancytopenia.

**Case Presentation:** We report on an eight-year-old boy who presented epistaxis and gum bleeding. The physical examination revealed petechiae, purpura, ecchymosis, and cervical while inguinal lymphadenopathy and splenomegaly were noted. *Brucella agglutinin* titer was positive. After five days of specific therapy for brucellosis, fever was controlled, clinical signs and symptoms were improved and platelet count was dramatically increased.

Conclusions: Sever thrombocytopenia and bleeding may be the presentation of brucellosis.

Keywords: Thrombocytopenia; Bleeding; Pediatric; Brucellosis

### 1. Introduction

Brucellosis is an important public health problem throughout the world, principally in the Mediterranean region, including Iran (1). The disease is transmitted to humans through contact with animals or their products. Consumption of fresh cheese, animal husbandry, working in a laboratory and having a veterinary profession are the main risk factors for brucellosis infection in Iran (2). Brucellosis symptoms are nonspecific; the most common complaints include fever, sweats, anorexia, headache, malaise, arthralgia, and lumbalgia (3). Hematological manifestations of active brucellosis vary from mild anemia and leukopenia to thrombocytopenia and rarely pancytopenia. The frequency of thrombocytopenia ranges from 3% to 20% (4). Hemorrhages have been reported in 3-19% of patients with brucellosis (3). Definite diagnosis of the infection is based on a positive blood or bone marrow culture for Brucella spp. While Wright agglutination could be a reliable test for the diagnosis of pediatric brucellosis (5). In this report, a case of brucellosis with severe thrombocytopenia and bleeding was presented.

## 2. Case Presentation

During April 2014, an eight-year-old boy was referred to the pediatric emergency unit of Afzalipour Hospital, Kerman University of Medical Sciences, Kerman, Southeastern Iran with epistaxis and gum bleeding. He had a history of fever, anorexia, and pallor since two months before admission and hematuria since four days before ago. History of unpasteurized dairy product consumption was not clear but the patient was from a rural area. On physical examination, petechiae, purpura, ecchymosis, cervical and inguinal lymphadenopathy and splenomegaly were noted.

For mucosal bleeding, local tranexamic acid was applied. Empirical antimicrobial therapy and IVig was started for treatment of severe bacterial infection and idiopathic thrombocytopenia on the basis of pediatric hematologist recommendation, yet after this therapy platelets count did not increase.

Laboratory tests results were as follows: white blood cell count 6800/mm<sup>3</sup>, with 25% lymphocytes and 75% neutrophils; platelets 14000/m<sup>3</sup>, hemoglobin 8.4 g/dL, erythrocyte sedimentation rate 100 mm/h, prothrombin time, 15.5 second: international normalized ratio, 1.22; and partial thromboplastin time, 34.4 second, lactate dehydrogenize 406 IU. Antinuclear antibody was negative. Blood urea nitrogen (BUN) and electrolytes were in normal ranges. Liver function tests were normal. Urine analysis showed microscopic. On the bone marrow aspiration (BMA) slide erythroid hyperplasia was noted without any evidence of malignancy or hemophagocytosis and leishman body was not detected. We did not perform bone marrow aspirate (BMA) flow cytometry. The abdominopelvic sonography did not show evidence of Para-aortic

Copyright @ 2016, Pediartric Infections Research Center; Published by Kowsar Corp.. This is an open-access article distributed under the terms of the Creative Commons Attribution-NonCommercial 4.0 International License (http://creativecommons.org/licenses/by-nc/4.0/) which permits copy and redistribute the material just in noncommercial usages, provided the original work is properly cited.

lymphadenopathy. Blood and bone marrow culture were negative for bacteria including Brucella. Serology tests for salmonella and Epstein-Barr virus (EBV) infection and also direct coombs test were negative. Brucella agglutinin titer was positive at 1:1024 and 2-mercaptoethanol test titer was positive at 1:320. On the basis of these results, brucellosis treatment was initiated with rifampin 15 mg/ kg/day, gentamicin 3 mg/kg/day, doxycycline 4 mg/kg/ day and other antimicrobial agents were discontinued. After five days, the fever was controlled, clinical signs and symptoms were improved and platelet count was dramatically increased. After 10 days he was discharged from hospital and therapy was continued with rifampin and doxycycline. At this time platelet count was 185000/ m3. In the second week of follow up, splenomegaly disappeared. History of unpasteurized goat's milk consumption was revealed yet screening of the patient's family did not detect any case of brucellosis.

## 3. Discussion

The frequency of thrombocytopenia in brucellosis is from 3% to 20% and is generally mild (4). In some cases, thrombocytopenia could be severe and symptomatic (6). Here, we report on a case in whom severe thrombocytopenia and bleeding was the presentation of brucellosis, which resolved promptly with specific triple anti-Brucella therapy. Triple therapy should be used in severe complicated cases (7). Thrombocytopenia in brucellosis could be due to hypersplenism, platelet sequestration and bone marrow suppression. In most patients who have thrombocytopenia, bone marrow is hypercellular (8). Hemophagocytic histiocytes in the bone marrow of patients with brucellosis as a cause of thrombocytopenia have been reported before (9), yet in this patient there were no hemophagocytic histiocytes in the marrow slide. An immune mechanism maybe another cause of platelet destruction, yet Coombs' test was negative in this patient (10). We could not show immune destruction of platelets by the usual tests, but it may be the responsible mechanism in this case. In conclusion, sever thrombocytopenia and bleeding may be the presentations of brucellosis. It is better for brucellosis to be included in the differential diagnosis when such presentations are observed, especially in endemic areas. Prompt diagnosis and treatment of this life threatening complication is essential.

## **Authors' Contributions**

Study concept and design: Ali Hosseininasab; interpretation of data: Ali Hosseininasab, Zahra Farahmandinia, Mahdieh Eslamian, Pegah Arfa, and Mothereh Abbaszadeh; critical revision of the manuscript for important intellectual content: Ali Hosseininasab, Zahra Farahmandinia, Mahdieh Eslamian, Pegah Arfa, and Mothereh Abbaszadeh.

## **Financial Disclosure**

We certify that the submission entitled "gum and nose bleeding as a presentation of pediatric brucellosis" has no conflict of interest including financial gain to report.

### References

- Sofian M, Aghakhani A, Velayati AA, Banifazl M, Eslamifar A, Ramezani A. Risk factors for human brucellosis in Iran: a case-control study. Int J Infect Dis. 2008;12(2):157–61.
- Zamani A, Kooraki S, Mohazab RA, Zamani N, Matloob R, Hayatbakhsh MR, et al. Epidemiological and clinical features of Brucella arthritis in 24 children. *Ann Saudi Med*. 2011;31(3):270–3.
- Baldane S, Sivgin S, Alkan TS, Kurnaz F, Pala C, Keklik M, et al. An atypical presentation of brucellosis in a patient with isolated thrombocytopenia complicated with upper gastrointestinal tract bleeding. *Case Rep Med*. 2012;**2012**:473784.
- Buzgan T, Karahocagil MK, Irmak H, Baran AI, Karsen H, Evirgen O, et al. Clinical manifestations and complications in 1028 cases of brucellosis: a retrospective evaluation and review of the literature. Int J Infect Dis. 2010;14(6):e469–78.
- Hosseininasab A, Alborzi A. Evaluation of Serum Agglutination Test in the Diagnosis of Pediatric Brucellosis. J Kerman Univ Med Sci. 2010;17(4):3.
- Citak EC, Citak FE, Tanyeri B, Arman D. Hematologic manifestations of brucellosis in children: 5 years experience of an anatolian center. J Pediatr Hematol Oncol. 2010;32(2):137–40.
- Alavi SM, Alavi L. Treatment of brucellosis: a systematic review of studies in recent twenty years. *Caspian J Intern Med.* 2013;4(2):636-41.
- Ulug M, Yaman Y, Yapici F, Can-Ulug N. Clinical and laboratory features, complications and treatment outcome of brucellosis in childhood and review of the literature. *Turk J Pediatr.* 2011;53(4):413–24.
- Erduran E, Makuloglu M, Mutlu M. A rare hematological manifestation of brucellosis: reactive hemophagocytic syndrome. J Microbiol Immunol Infect. 2010;43(2):159–62.
- Farah RA, Hage P, Al Rifai A, Afif C. Immune thrombocytopenic purpura associated with brucellosis. Case report and review of the literature. J Med Liban. 2010;58(4):241-3.