Case Report

A rare presentation of peripheral edema and ascites in a 10-year-old child with brucellosis: A case report

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ABSTRACT

Introduction: and importance: Brucellosis is a common infection in Mediterranean region that manifests with various symptoms. Brucellosis should be considered as a possible cause of recurrent fever even if the symptoms are not suggestive of brucellosis.

Case presentation: We report a case of 10-year-old child with no significant past medical history who presented with a 4-day period peripheral edema and ascites without fever, arthralgia or abdominal pain.

Clinical discussion: Proper investigations showed normal cardiac and renal functions; ultrasonography showed no portal vein hypertension. Albumin and total protein were also within normal. Complete blood count revealed pancytopenia; bone marrow aspiration and biopsy revealed hypercellularity that could be attributed to hypersplenism as a possible cause. Liver biopsy revealed non-specific inflammatory findings and also did not lead to a definite diagnosis. While broadening the scope of deferential diagnosis in order to reach a final diagnosis, Wright serum agglutination was tested positive (1/640) and we diagnosed a brucellosis infection. A proper management with Antibiotics ensued; the patient had uneventful recovery on treatment until complete clinical and imaging resolution of signs and symptoms.

Conclusion: Although brucellosis is considered a multi-systemic disease with atypical presentations, early diagnosis of brucellosis with management causes rapid recovery and favorable prognosis. We report a case of ascites and edema in context of Brucella infection which was completely resolved after treatment. This condition is rare especially in previously healthy child and after excluding other possible causes. We aim to share our case to keep brucellosis in mind as a differential diagnosis when dealing with infectious diseases with non-specific symptoms.

1. Introduction

Brucellosis is a febrile disease with insidious onset and various manifestations. Its clinical presentation (fever, headache, myalgia, malaise, night sweats and weight loss) can mimic many other diseases. This infection is considered a multi-system disease; which might affect the alimentary tract, locomotor, respiratory, cardiovascular, central nervous and genitourinary system [1]. Atypical presentations such as ascites and peripheral edema may also occur seldom. Few cases in the medical literature reported an association between ascites and brucellosis [2,3]; even fewer mentioned peripheral edema emerging in the context of brucellosis [4]. Most cases found in the literature attributed ascites to a specific underlying etiology such as liver disease or spontaneous bacterial peritonitis [3,5,6], whereas peripheral edema rarely accompanied capillary leak syndrome [4,7].

However, an association between ascites, peripheral edema and brucellosis without an evident mechanism was not found in the literature. In this article, we report a case of brucellosis that manifested with ascites and peripheral edema, posing a challenging diagnosis on the clinician.

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2. Case presentation

A 10-year-old child with no significant past medical and family history presented to the pediatric emergency department of Aleppo University Hospital complaining of a 4-day period peripheral edema over both lower extremities and ascites without fever, arthralgia or abdominal pain. The child had a history of jaundice, dark urine and abdominal pain 16 days ago which resolved spontaneously suggesting acute hepatitis.

At time of admission, the patient general appearance was well. Upon physical examination, mild pallor, tachycardia, tachypnea, palpable soft hepatomegaly 6 cm below the right costal margin, palpable splenomegaly 8 cm below the left costal margin, ascites, skin cracks, venous graftiti on the chest wall, severe pitting edema on the lower extremities, petechiae on the thighs were found. While admitted at the hospital, the patient developed a recurrent fever (39–39.5 °C) without abdominal pain or arthralgia. Complete blood count at admission revealed anemia (Hb: 6.6 g/dL), leukopenia (WBC: 3,800/mm3), thrombocytopenia (PLT: 18,000). The evaluation of the blood film revealed: RBCs (severe normocytosis, severe anisocytosis, microcytic, hypochromic anemia), severe poikilocytosis (many ring cells, schistocytes, and spherocytes) - WBCs (slight leukopenia, neutrophil differential shift to the left, and immature forms (band cells) - PLTs (severe thrombocytopenia) - reticulocyte count 6%. Na, K, Kidney Function tests, Glucose were within normal limits.

Abdominal ultrasonography revealed homogeneous enlargement of the liver, increased flow rate in the portal vein without any signs of thrombosis, homogeneous enlargement of the spleen with congestion in the vessels of the splenic umbilicus, and ascites of a large amount. As acute hepatitis was suggested by the clinical presentation, we ran HAV IgM at the time of admission but the result was negative. Considering other causes of hepatitis, ANA, ASMA, Anti-LKM were performed, but the results came back negative as well. The evaluation of bone marrow aspirate revealed hypercellularity which might be attributed to hyper-splenism. Moving on with the diagnostic process, we performed a liver biopsy. Biopsy showed non-specific chronic hepatitis with mild congestion. All previous investigations were not conclusive. Therefore, we needed further testing for possible differential diagnoses. Vidal test for salmonella was negative. The diagnosis of brucellosis was established by a positive Wright serum agglutination test for Brucella (1/640).

We initiated treatment with proper antibiotics immediately; IV Gentamycin 48 mg tid, PO Rifampin 10 mg/kg/day bid and PO Doxycycline 30 mg/kg/day qid for 6 days. The symptoms and signs receded gradually, and the patient was discharged on a 5-week Rifampin and Doxycycline prescription. Subsequent follow-up abdominal ultrasonography showed normal homogeneous liver, normal homogeneous spleen and low amount of free abdominal fluid. Flow rate of the portal vein was normal. After completion of treatment, the patient recovered completely and all symptoms resolved.

3. Discussion

Brucellosis is a systemic disease that may involve any organ or tissue caused by small, fastidious gram-negative cocobacilli [8]. Common sources of infection are raw milk, cheese, meat, or through direct contact with infected animals, or animal excreta. Our patient had no contact with any farm or wild animals which makes it more likely to be caused by digestion of contaminated foods. The exact incidence of human brucellosis is still unknown [9]. Human brucellosis has a wide spectrum of symptoms, the most prominent symptoms of brucellosis are fever, sweats, malaise, anorexia, arthralgia and back pain [10].

Few data on the frequency of edema and ascites occurring in brucellosis especially in children have been reported, while there have been reports of hepatic abscess [11], acute abdomen [12], enteritis [13] cholecystitis [14], and pancreatitis [15] attributed to Brucella infection. In this manuscript, we report a child with brucellosis presenting with peripheral edema and ascites without the common constitutional symptoms. The presence of ascites is an extremely rare finding in the context of brucellosis, often related to certain conditions like underlying liver disease [3]. In such cases, ascites was mostly attributed to portal hypertension, hypoalbuminemia or simply to direct peritoneal infection with Brucella [5,6].

In other cases; where patients were previously healthy and no predisposing conditions were marked, different etiologies were proposed to explain ascites. Some studies suggested that ascites was a result of peritonitis related to vasculitis in the context of mixed cryoglobulinemia triggered by brucellosis [16,17]. Akritidis N et al. attributed ascites to a generalized abdominal immune reaction to Brucella [3]. The latter explanation is the most convenient to our case, as the investigations were not indicative of a predisposing liver disease, portal hypertension or vasculitis.

Peripheral edema is an important manifestation of many diseases. However, it is rarely a manifestation of brucellosis, very few studies have mentioned edema as a prominent symptom in brucellosis, most of them proposed low albumin levels as a mechanism of formation of edema.

Another mechanism was described in medical literature, two studies reported edema in the course of capillary leak syndrome (CLS) precipitated by Brucellosis [4,7]. CLS is an unusual condition characterized by unexplained capillary hyperpermeability commonly following an inflammatory stimulus such as viral or bacterial infections. It manifests with a localized or diffuse edema, preceded by fever, and hypotension, and is usually associated with hypoproteinemia, hypoalbuminemia and IgG type monoclonal gammopathy.

In our case, Peripheral pitting edema was clinically apparent over the lower extremities. Physical examination and laboratory tests ruled out cardiac, hepatic and renal causes of peripheral edema and normal albumin and total protein ruled out CLS. Therefore, none of the causes of edema mentioned in the literature were similar to our case.

Brucellosis causes a diversity of nonspecific hematologic abnormal findings. Hematologic complications such as mild anemia and leukopenia have been frequently associated with acute brucellosis but pancytopenia is less frequently seen [18].

Hemophagocytosis, hypersplenism, immune destruction or granulomatous changes in bone marrow may be responsible for pancytopenia in brucellosis [19].

In our case the patient had pancytopenia with hypercellularity on bone marrow examination, which indicates hypersplenism as the probable cause. Our findings indicated that pancytopenia was not due to bone marrow failure or malignancy, and pancytopenia completely resolved after treatment.

Concluding, the infection with Brucella could be expressed in a variety of clinical symptoms, which might be non-specific or misleading; therefore, Brucellosis should always be a diagnostic possibility and should be kept in the mind of the clinician.

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Author contribution

BS and MA contributed to the conception and design of work. LZ and BW contributed to data collection. BS and RB and MC contributed to the critical revision of the article. All authors read and approved the final manuscript.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2021.103196.

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