

A Rare Manifestation of Brucellosis: Cervical Lymphadenitis

Bruselozisin Nadir Klinik Prezentasyonu: Servikal Lenfadenit

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Abstract

Human brucellosis is a multisystem and potentially lethal disease of zoonotic origin with highly variable and nonspecific clinical presentation. Although lymphadenopathy in the course of brucellosis is 10-20%, there is only one case of isolated cervical LAP as unique manifestation. We hereby present a case of isolated cervical LAP due to brucella mellitensis in a 12 year old boy.

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Key words: Brucellosis, lymphadenopathy

Özet

Bruselozis, oldukça değişken ve özgül olmayan bulgularla kliniği yansayan, bütün sistemleri tutabilen, hayvan kaynaklı, öldürücü potansiyele sahip bir hastalıktır. Lenfadenopati, brucellosis'in seyri esnasında %10-20 vakada görülmekte birlikte, izole servikal lenfadenit ile başvurulan sadece bir vaka vardır. Burada, izole servikal lenfadenit ile başvuran oniki yaşındaki bir erkek çocuk sunulacaktır.

(*Çocuk Enf Derg* 2009; 3: 190-1)

Anahtar kelimeler: Bruselozis, lenfadenopati

Introduction

Human brucellosis is a multisystemic and potentially lethal disease of zoonotic origin with highly variable and nonspecific clinical presentations (1). Although the occurrence of lymphadenopathy (LAP) in the course of brucellosis is 10-20%(2), there is only one case of isolated cervical LAP as a unique manifestation. Hereby in this study, a case of isolated cervical LAP due to brucella mellitensis in a 12 year-old boy is presented.

Patient Report

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consumption of unpasteurized dairy products. On admission, he was febrile (38.5°C) and tachycardic (124 beats /min) and a cervical lymph node enlargement, 8x8 cm in size in the left anterior region, was detected.

There was no rash, organomegaly or tenderness over the bones and no evidence of joint swelling or effusion. Neurological examination and other physical examinations did not reveal any significant pathology.

Laboratory findings on admission were as follows: haemoglobin level of 11.8 gr/dl, leukocyte count of $18.9 \times 10^9 /L$, platelet count of $310 \times 10^9 /L$, erythrocyte sedimentation rate of 52 mm/h and C-reactive protein was 9.7 mg / dl. Renal and liver function tests were within normal limits. On peripheral blood smear, 70%Polymorphonuclear leucocytes, 20%lymphocytes, 10%monocytes were observed and no atypical lymphocytes were reported. Chest X-ray was normal. Considering the mentioned results, he was diagnosed as cervical suppurative lymphadenitis. Incision was performed for drainage and a pus specimen was sent to the laboratory for routine cultures such as aerobic, anaerobic and mycobacterium specific culture.

Cefazolin and amikacin therapy was started empirically. Unexpectedly, no organism from cultures was detected. Gram stain and Ziehl-Nelson stain were all negative. Attempts to isolate any infecting organisms from the blood were unsuccessful. On the 11th day of treatment, right cervical lymph node enlargement and fluctuancy were reported. There was no decrease in size of the left sided lymphadenopathy and the subsided fever increased again. Repeated blood cultures revealed no bacterial growth. To rule out any malignancy, excisional lymph node biopsy, bone marrow examination, Computed tomography (CT) scan of abdomen, chest and neck were done. CT of the neck showed conglomerate lymphadenopathy on each side of the neck some of which had a necrotizing nature (Fig 1). Excisional lymph node biopsy showed nonspecific reactive hyperplasia and acid fast bacilli on Ziehl Nelson staining was negative.

Viral serology for Ebstein Barr Virus, Cytomegalovirus, Toxoplasmosis, Human Immune Deficiency Virus and Hepatitis virus were all negative. Tuberculin skin test and nitroblue tetra zolum dye reduction tests were also negative. Thus the patient had received a course of amikacin for 11 days and Cefazolin for 21 days. At the end of the third week, the report of Brucella agglutination test was received showing 1: 1280 titers, Elisa IgM positive and IgG negative. Doxycycline, streptomycin and rifampicin treatment was started. Streptomycin therapy was stopped on the 15th day.

The patient improved very slowly with the drug treatment. Even though the patient had gained 3 kg, he still had a draining sinus from incised lymphadenopathy at the end of sixth week. Therefore duration of therapy was extended to 12 weeks. After completion of the treatment the patient recovered with no LAP and brucella agglutination titer decreased to 1: 640. After 6 months of follow up, no relapse was reported and the patient had gained 5 kg weight during this period.

Discussion

Brucellosis is a systemic disease that can involve any organ or system of the body. Human brucellosis is known for protean manifestations (3, 4) such as, joint pain, low backache, night sweats, cough, testicular pain, anorexia, jaundice, headache, fatigue, convulsions, hepatosplenomegaly, weight loss, swollen hands, and burning feet. Although the most

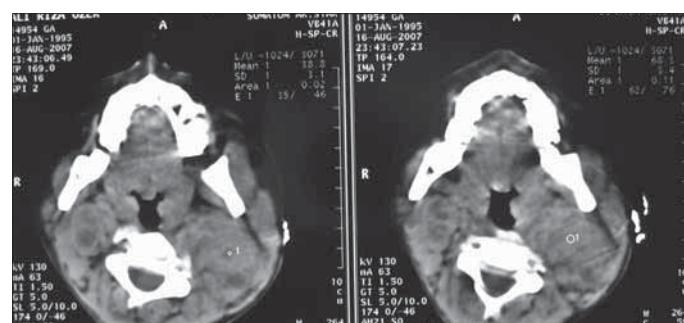


Figure 1. CT of neck showing lymphadenopathy on each side of neck with necrotizing nature on the right side

frequent form of brucella is a systemic one, local forms of brucella can occur in bones, joints, heart, lung, central nervous system, liver, spleen, testes and the ovaries (5, 6). Lymphadenopathy can be seen in 3.1-20% of patients (7, 2). Nevertheless brucellosis with localized lymphadenitis has been rarely reported (8, 6). Nadler et al. reported a 54 year old woman with suppurative lymphadenitis accompanied by erythema, pain, fever, and pus drainage. Brucella suis was detected as a causative agent. Varane et al. reported 42 year old male with an isolated cervical lymphadenopathy due to Brucella melitensis without systemic signs and symptoms.

In the above presented case, even though no brucella species from obtained sample (blood cultures, pus) was detected, the positive results of brucella standard tube agglutination test (STA) and Elisa for brucella were considered and the treatment with streptomycin, doxycycline and rifampicin was started. Blood culture provides definite proof of brucellosis but may not provide a positive result for all patients even under ideal conditions (2). Antigen detection by enzyme-linked immunosorbent assay as an alternative to blood culture is reported (9). Even though no brucella species on blood culture was detected, in this presented case both Elisa and STA being positive denoted diagnosis for brucella. With proper treatment the patient improved and at the follow up during sixth months no relapse was detected.

To the best of our knowledge, this is the first report of lymphadenitis associated with brucella in childhood. It is proposed that, in the differential diagnosis of localized lymphadenitis, brucellosis should be kept in mind.

References

- Lee MK, Fung KS. A case of human brucellosis in Hong Kong. *Hong Kong Med J* 2005; 11: 403-6.
- Colmenero JD, Reguera JM, Martos F, Sanchez-De-Mora D, Delgado M, Causse M, Martin-Farfán A, Juárez C. Complications associated with *Brucella melitensis* infection: A study of 530 cases. *Medicine (Baltimore)* 1996; 75: 195-211. [Erratum, in: *Medicine (Baltimore)* 1997; 76: 139].
- Mantur BG, Biradar MS, Bidri RC, Mulimani MS, Veerappa, Kariholu P, Patil SB, Mangalgi SS. Protean clinical manifestations and diagnostic challenges of human brucellosis in adults: 16 years' experience in an endemic area. *J Med Microbiol* 2006; 55: 897-903.
- Mantur BG, Akki AS, Mangalgi SS, Patil SV, Gobbur RH, Peerapur BV. Childhood brucellosis - a microbiological, epidemiological and clinical study. *J Trop Pediatr* 2004; 50: 153-7.
- Fenkci V, Ceviroglu S, Yilmazer M. Ovarian abscess due to *Brucella melitensis*. *Scand J Infect Dis* 2003; 35: 762-3.
- Varona JF, Guerra JM, Guillén V, Guillén S, Menassa A, Palenque E. Isolated cervical lymphadenopathy as unique manifestation of Brucellosis. *Scand J Infect Dis* 2002; 34: 538-40.
- Mantur BG, Amarnath SK, Shinde RS. Review of clinical and laboratory features of human Brucellosis. *Indian J Med Microbiol* 2007; 25: 188-202.
- Nadler H, Dolan C, Forgacs P, George H. *Brucella suis*: an unusual cause of suppurative lymphadenitis in an outpatient. *J Clin Microbiol* 1982; 16: 575-6.
- Al-Shamahy HA, Wright SG. Enzyme-linked immunosorbent assay for *Brucella* antigen detection in human sera. *J Med Microbiol* 1998; 47: 169-72.