

Neurobrucellosis with Gait disturbance: A Neurological case report

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Abstract

Brucellosis is a multi-system infectious disease that exhibits with various manifestations and complications. Neurobrucellosis is a rare but serious presentation of brucellosis that can be discovered in every stages of the disease. Laboratory tests and physical examination and patient history are generally the basis for diagnosing the disease. It has both insidious and prolonged clinical course of the disease and long-term therapies. Also the most common pattern of exhibition is subacute or chronic. We reported a case of young female who had history of painless weakness in the right lower limb (proximal and distal) that started gradually and had progressed over the time, and after a month she felt weakness in the left lower limb with the same pattern. Lumbosacral Magnetic resonance imaging (MRI) with and

without contrast was shown evidence of enhancement thickening of caudal equina ventral roots. Brucella antigen titer was positive, the result was 1/160. And other clinical tests were normal.

Patient treated with Intravenous injection (IV) Rifampicin and Intravenous Cotrimoxazole. patient was discharged with good health and continuing all two medications for 5 months.

The decision was taken to report this case as a result of entire respond in patient's illness after a enduring disease. Neurobrucellosis is a treatable disease in which it would be better to consider a high indication of suspicion. Due to if ignored, it may cause significant morbidity and mortality.

Keywords: Neurobrucellosis, gait disturbance, acid paraparesis, weakness

Introduction

The most frequent bacterial zoonosis disease is Brucellosis and lead to more than 500,000 human infections per year worldwide.[1] Although, the disease has been reported all over the world, it has a higher widespread in countries where the health care problems and animal health are not standardized. In Asian countries and Turkey, a high prevalence of the disease has been reported.[2][3]

Different and non-specific clinical manifestation make the diagnosis difficult. The most frequent symptoms are fever, myalgia, arthralgia, weight loss and night sweats.[2] Neurologic findings of brucellosis occur in less than 5% in adolescents[4][5][6] but its incidence in pediatric is less than 0.8-1%.[7][8]

Neurological complications include encephalitis, meningoencephalitis, radiculitis, myelitis, peripheral and cranial neuropathies, subarachnoid hemorrhage, psychiatric manifestations, brain abscess and demyelinating syndrome.[9][10]

In endemic areas, if patient is discovered with neurological symptoms, diagnostic tests including: serum antibodies detection, isolation of cerebrospinal fluid (CSF), and bone marrow must be performed. Examining the patient's complete history such as travel history, occupation and similar symptoms in other family members could also be very helpful to rule out neurobrucellosis.[11] Analysis of CSF reveals an elevated protein concentration, moderate leukocytosis and hypoglycorrhachia.[11][12] CSF and blood cultures can be negative. Thus, the diagnosis is made by detecting brucella antibodies in CSF. This is

diagnostic.[13]

We report an 13-year-old female patient with painless weakness in the both lower limb with same patterns, gait disturbance, accid paraprasia and loss of appetite. That has rarely been reported as a manifestation of neurobru-cellosis.

Case Report

A 13-year-old female patient was admitted in August in Firoozgar hospital, with history of gait distrurbance and loss of appetite without weight loss for 3 months. She'd had painless weakness in the right lower limb (proximal and distal) that started gradually and had progressed over the time, and after a month she felt weakness in the left lower limb with the same pattern. On arrival in the hospital, she had no fever and other parts of the vital signs were normal.No history of vomiting, headache, other sensorium, or seizure was reported. The patient belonged to a rural area and There is no history of contact with cows and goats and no raw milk has been consumed. Her vaccination program during infancy and childhood had been completely done and she had not any recent vaccination.Her parents were not relatives. Her father had treated brucellosis 20 years ago and her mother had been on treatment until 3 month ago. She had two younger sister and neither of them has the same symptoms.

On nervous system examination, our patient was conscious, Motor examination revealed normal muscle bulk, grade III power in proximal lower limb and grade II in distal and proximal force of lower limb. power of upper limb was normal. Deep Tendon Re ex (DTR) of upper limb was 2+ , ankle and knee were 0. Plantar response was donward bilatrally.Laboratory tests including Complete Blood Counts (CBC), Blood Urea Nitrogen (BUN), serum creati-nine, electrolytes and liver function tests proved to be normal. Erythrocyte Sedimentation Rate (ESR) was 6 and C-Reactive Protein (CRP) 1. SSA-RO and SSB-LA both were 0.1 . Antibody for HIV virus, HBs Antigen and Anti HCV were negative. Lumbar puncture was performed and checked for anal-ysis (Table 1). Serological test results were positive(Table 2). Lumbosacral MRI with and without contrast was shown evidence of enhancement thicken-ing of caudal equina ventral roots (Fig.1).Because of normal Sensory Nerve Action Potential (SNAP) and decrease amplitude of Compound Muscle Ac-tion Potential(CMAP), the result of Electromyography (EMG) has shown subacute involvement of Anterior Horn Cell, especially in L5, S1 territory.

Treatment was commence with Intravenous injection (IV) Rifampicin and In-travenous Cotrimoxazole. patient was discharged with good health and con-tinuing all two medications (Rifampicin and Cotrimoxazole) for 5 months. She returned to hospital with recurrence symptoms due to discontinue med-ications before complete the course of treatment.

Fluid Bodies	
Test	Result
COMPLETE CSF	
CSF	.
W.B.C	150
R.B.C	0
Glucose	19
Total Protein	152
LDH	
LDH-P	53
Di Result	
Segment	20
Lymphocyte	80

Table 1: Pattern of CSF analysis

Serology			
Test	Result	Unit	References
Wright	1/160	Titer	Positive: >= 1/80
Coombs Wright	1/160	Titer	Positive: >= 1/80
2ME ₂ Wright(IgG)Titer	1/80	Titer	Positive: >= 1/40
PPD			
Nodular reaction	Borderline		
State	7 mm	mm	Negative: <5 mm Borderline : 5-10 mm Positive : >10 mm

Table 2: Serological ndings

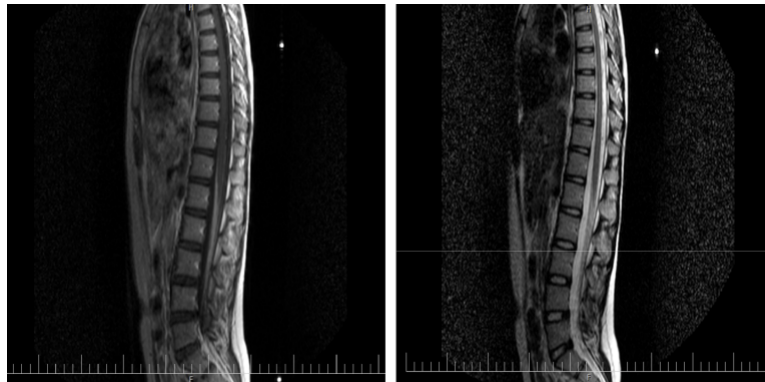


Figure 1: enhancement of caudal equina ventral roots

Discussion

It was observed that neurobrucellosis rarely reported on reviewing literature. Brucellosis is an endemic infectious disease in Iran. [14] Neurobrucellosis accounts for about 3-5% of cases of brucellosis.[11] The most common forms of neurobrucellosis are Meningitis and meningoencephalitis.[12] we reported a patient with gait disturbance, which is rare among reported cases.

The consumption of unpasteurized milk products is one of the most common cases of transmission of the disease.[15] yet our case, despite being rural, has no history of consumption of unpasteurized milk. Also Our patient presented with enhancement of caudal equina ventral roots in MR imaging and positive serology test. In one study which carried out in Saudi Arabia, they found clinical manifestations were related to the imaging abnormalities. They also categorized the changes of nervous system involvement of neurobrucellosis into 4 categories including: 1) normal, 2) inflammation (abnormal enhancement), 3) alteration of white matter, 4) and radiologic findings which display vascular alteration.[16]

In another study, Gul HC, Erdem H, Gorenk L et al. from Turkey reported that the diagnosis was established by serum antibodies test and CSF findings in 11 cases of neurobrucellosis. And they did not utilize imaging techniques

to diagnose it.[17]

In treatment of this disease, due to the central nervous system is involved, antibiotics are used that are able to cross the blood-brain barrier well. It is preferable to use Co-trimoxazole, Doxycycline and Rifampicin. Combination treatment with at least two medications of those indicated earlier is recommended.[2][12][18][18] The decision was taken to use Rifampicin and Co-trimoxazole for this patient.

In conclusion, Neurobrucellosis as an infectious disease, can be treated with a favorable result. The diagnosis of this disease is contingent generally upon high clinical attention in endemic areas such as Iran. The disease differs in clinical diagnosis or radiology, especially in young patients with neurological disorders. Imaging neurobrucellosis results can be potentially misleading because they are different and can mimic other infectious, or inflammatory diseases like Tuberculosis. The sentence of what follows is drawn to the literature Harrison's textbook of Medicine. Patients should ideally be followed clinically for up to 2 years because recurrence occurs in up to 30% of patients.

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References

- [1] Georgios Pappas, Photini Papadimitriou, Nikolaos Akritidis, Leonidas Christou, and Epameinondas V Tsianos. The new global map of human brucellosis. The Lancet infectious diseases, 6(2):91{99, 2006.
- [2] Hane Cem Gul, Hakan Erdem, and Semai Bek. Overview of neurobrucellosis: a pooled analysis of 187 cases. International Journal of Infectious Diseases, 13(6):e339{e343, 2009.
- [3] Osman Kizilkilic and Cem Calli. Neurobrucellosis. Neuroimaging Clinics, 21(4):927{937, 2011.

- [4] EJ Young, GL Mandell, JE Bennett, and R Dolin. *Brucella species principles and practice of infectious diseases*. Churchil livingstone, 226, 2009.
- [5] Donald R McLean, Neville Russell, and M Yousuf Khan. Neurobrucellosis: clinical and therapeutic features. Clinical infectious diseases, 15(4):582{590, 1992.
- [6] Gerald L Mandell, RG Douglas Jr, John E Bennett, et al. Principles and practice of infectious diseases. volumes 1 and 2. Principles and practice of infectious diseases. Volumes 1 and 2., 1979.
- [7] YKR Habeeb, AKN Al-Najdi, SAH Sadek, and E Al-Onaizi. Paediatric neurobrucellosis: case report and literature review. Journal of infection, 37(1):59{62, 1998.
- [8] Aira Bucher, Peter Gaustad, and Eivind Pape. Chronic neurobrucellosis due to brucella melitensis. Scandinavian journal of infectious diseases, 22(2):223{226, 1990.
- [9] N Akritidis, M Bosilkovski, and G Pappas. Medical progress brucellosis. The New En. J. Med, 352:2325{2336, 2005.
- [10] Tumer Guven, Kenan Ugurlu, Onder Ergonul, Aysel Kocagul Celikbas, Sebnem Eren Gok, Selcuk Comoglu, Nurcan Baykam, and Basak Dokuzoguz. Neurobrucellosis: clinical and diagnostic features. Clinical Infectious Diseases, 56(10):1407{1412, 2013.
- [11] Raad A Shakir, ASN Al-Din, GF Araj, AR Lulu, AR Mousa, and MA Saadah. Clinical categories of neurobrucellosis: a report on 19 cases. Brain, 110(1):213{223, 1987.
- [12] Mahboubah Haji-Abdolbagi, Mehrnaz Rasooli-Nejad, Sirous Jafari, Mehrdad Hasibi, and Abdolreza Soudbakhsh. Clinical and laboratory findings in neurobrucellosis: review of 31 cases. Arch Iran Med, 11(1):21{5, 2008.
- [13] Daniel Tena, Alejandro Gonzalez-Praetorius, Alberto Lopez-Alonso, Jose Luis Peña, María Teresa Perez-Pomata, and Julia Bisquert. Acute meningitis due to brucella spp. European journal of pediatrics, 165(10):726, 2006.
- [14] Saleh M Al Deeb, Basim A Yaqub, Hassan S Sharif, and Jayent G Phadke. Neurobrucellosis: clinical characteristics, diagnosis, and out-come. Neurology, 39(4):498{498, 1989.

- [15] M Turgut, AT Turgut, and U Kosar. Spinal brucellosis: Turkish experience based on 452 cases published during the last century. Acta neurochirurgica, 148(10):1033{1044, 2006.
- [16] M Walid Al-Sous, Saeed Bohlega, M Zuheir Al-Kawi, Jehad Alwatban, and Donald R McLean. Neurobrucellosis: clinical and neuroimaging correlation. American Journal of Neuroradiology, 25(3):395{401, 2004.
- [17] Hane Cem Gul, Hakan Erdem, Levent Gorenek, Mehmet Fatih Ozdag, Yasin Kalpakci, Ismail Yasar Avci, Bulent Ahmet Besirbellioglu, and Can Polat Eyigun. Management of neurobrucellosis: an assessment of 11 cases. Internal Medicine, 47(11):995{1001, 2008.
- [18] J Bosch, J Linares, MJ Lopez de Goicoechea, J Ariza, MC Cissal, and R Martin. In-vitro activity of cipro oxacin, ceftriaxone and ve other antimicrobial agents against 95 strains of brucella melitensis. Journal of Antimicrobial Chemotherapy, 17(4):459{461, 1986.