

Encephalomyelitis Due to Brucellosis

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Introduction: Human brucellosis is a public health problem worldwide. Neurobrucellosis is a rare and severe form of systemic infection and has a broad range of clinical syndromes.

Case Presentation: We reported a 21-year-old woman who was referred to our clinic with a subacute onset of fever, myalgia, and weakness of right foot, which gradually progressed and involved lower limbs and culminated in total paralysis.

Conclusions: Although neurobrucellosis is a rare presentation of disease, meningitis or meningoencephalitis are the most frequent clinical syndromes associated with this form of infection. Brucellosis should be considered in patients with unexplained neurological findings, particularly in brucellosis endemic area.

Keywords: Brucellosis; Neurobrucellosis; Meningoencephalitis

1. Introduction

Brucellosis is an extremely important disease worldwide, especially in developing countries such as Iran. Its clinical manifestations and severity vary according to the *Brucella* species and involved organ. Men aged 13 to 40 years are particularly susceptible to the disease due to *Brucella* species. Possible explanation includes activities that increase exposure to *Brucella* species (1, 2). Fever is the most common symptom. Constitutional symptoms including anorexia, malaise, fatigue, weakness, and weight loss are very common. Other common clinical symptoms include low back pain, joint pain, and rarely, joint swelling. Neurobrucellosis is a rare and severe form of systemic infection. The most frequent central nervous system (CNS) infection-associated clinical syndromes are meningitis or meningoencephalitis (2-4). Encephalomyelitis is a general term for inflammation of the brain and spinal cord, describing a number of disorders and acute disseminated encephalomyelitis. Multiple sclerosis (MS) is a demyelinating disease of the CNS, which is possibly triggered by infection, environmental factors, stress, smoking, autoimmune disorders, and genetic factor (5-9). Here, we reported a 21-year-old woman who was referred to our clinic with a subacute onset of fever, myalgia, and weakness of right foot, which gradually progressed to total paralysis.

2. Case Presentation

The patient was a 21-year-old woman who was referred to our clinic with a subacute onset of fever, myalgia, and

weakness of right foot, which gradually progressed to involve lower limbs and culminated in total paralysis. She was treated for depression and lack of sleep during the preceding month. Nystagmus and cerebellar ataxia were other manifestations. On admission day, her oral temperature was 38.2 °C and blood pressure was 110/50 mm Hg. Her pulse and respiratory rates were 85 and 19 per minutes, respectively. The findings of cardiac and pulmonary examinations were insignificant. No other abnormal signs were observed. A complete blood count revealed a white blood cell count of $11.2 \times 10^9/L$ (neutrophils, 52%; and lymphocytes, 35%), hemoglobin of 12.8 g/dL, and a platelet count of $230 \times 10^9/L$. Blood urea nitrogen and creatinine levels were within normal limits. Blood glucose was 97 mg/dL. Chest X-ray findings were insignificant. Magnetic resonance imaging (MRI) characteristically showed the lesions of variable location in the white matter of the brain, and enhancement along the distal cord in lumbar and cauda equina. Cerebrospinal fluid (CSF) showed a xanthochromia with elevated protein level, lymphocytic pleocytosis, and low sugar (45 mg/dL). She was treated with ceftriaxone, vancomycin, and acyclovir. CSF sample and blood sample were examined for brucella, because she was from Khash, a city in Sistan and Baluchistan province, where brucellosis has the highest prevalence rate among other cities of the province. Wright test in blood and CSF revealed positive results at 1:320 and 1:40 titers, respectively. The same results were achieved by 2-mecaptoethanol at 1:160. Neurologic consultation approved the diagnosis of MS, possibly due to brucellosis. We changed the drug regimen to 4 g/

day of intravenous ceftriaxone, 200 mg/day of oral doxycycline, and 600 mg/day of rifampin. In addition, high-dose steroid was started. Twenty-five days later, she was discharged and referred to neurology center.

3. Discussion

Brucellosis is a zoonotic infection caused by the bacterial genus *Brucella*. Humans are accidental hosts, but brucellosis continues to be an important public health problem worldwide, which reflects spreading of disease in animals (1, 2). Fever is the most common symptom. It is associated with chills in almost 80% of patients. Constitutional symptoms of brucellosis are very common (> 90%) and include anorexia, malaise, fatigue, weakness, and weight loss. Other common clinical symptoms include low back pain, joint pain, and rarely, joint swelling. These symptoms affect as many as 50% to 80% of patients. Arthralgias may be diffuse or localized, with a predilection for epiphysis and the sacroiliac joint (1-3). Brucellosis should be considered in any patient whose place of residence, dietary, and occupational history suggests a risk for the infection and in those who are experiencing any of the various known neurological or non-neurological complications in brucellosis endemic areas. Neurobrucellosis is a rare and severe form of systemic infection and has a broad range of clinical syndromes. The most frequent clinical syndromes associated with brucellosis are meningitis or meningoencephalitis (2-4). Encephalomyelitis is a general term for inflammation of the CNS, describing a number of disorders and acute disseminated encephalomyelitis. MS is a demyelinating disease of the CNS, which is possibly triggered by infection (mostly viral), stress, smoking, autoimmune disorders, and genetic factor (5-9). A patient with MS can have almost any neurological sign or symptom including motor, sensory, and visual problems, which are the most common presentation. The specific symptoms are related to the locations of the lesions within CNS. These symptoms may be loss of sensitivity or changes in sensation such as pins and needles or numbness, muscle spasms, muscle weakness, or difficulty in moving and coordination. Visual problems such as nystagmus, optic neuritis, or double vision can be seen (5, 6). Many rare complications of neurobrucellosis have been reported (2-4, 10). Yilmaz et al. reported

a case of neurobrucellosis with hydrocephaly (2). Tugcu et al. reported a case of chronic meningitis due to brucellosis leading to complicated intracranial hypertension (3). Bektas et al. presented a case of neurobrucellosis that similar to our patient, presented as demyelination disorder (4). Sometimes neurobrucellosis complication can present as an acute disseminated encephalomyelitis. On the other hand, some of the signs and symptoms of brucellosis can mimic MS manifestations. This condition in our patient could be a comorbidity of brucellosis and MS due to another cause or MS can present as a systemic complication of brucellosis. Our patient responded to antimicrobial agents. However, according to neurology consultation, we started high-dose steroid too. We conclude that brucellosis should be considered in any patient who is experiencing any of the various known neurologic or complications of brucellosis in endemic areas.

Authors' Contributions

Mohammad Hashemi-Shahri, Maliheh Metanat, and Maliheh Kooshki wrote the manuscript.

References

1. Keshtkar-Jahromi M, Razavi SM, Gholamin S, Keshtkar-Jahromi M, Hossain M, Sajadi MM. Medical versus medical and surgical treatment for brucella endocarditis. *Ann Thorac Surg*. 2012;**94**(6):2141-6.
2. Yilmaz C, Kaya A, Guven AS, Yilmaz N, Caksen H. A case of neurobrucellosis with hydrocephaly. *West Indian Med J*. 2013;**62**(7):678-9.
3. Tugcu B, Nacaroglu SA, Coskun C, Kuscu DY, Onder F. Chronic Meningitis Complicating Intracranial Hypertension in Neurobrucellosis: A Case Report. *Semin Ophthalmol*. 2014.
4. Bektas O, Ozdemir H, Yilmaz A, Fitoz S, Ciftci E, Ince E, et al. An unusual case of neurobrucellosis presenting as demyelination disorder. *Turk J Pediatr*. 2013;**55**(2):210-3.
5. Nakahara J, Maeda M, Aiso S, Suzuki N. Current concepts in multiple sclerosis: autoimmunity versus oligodendroglialopathy. *Clin Rev Allergy Immunol*. 2012;**42**(1):26-34.
6. Tsang BK, Macdonell R. Multiple sclerosis- diagnosis, management and prognosis. *Aust Fam Physician*. 2011;**40**(12):948-55.
7. Hassan-Smith G, Douglas MR. Epidemiology and diagnosis of multiple sclerosis. *Br J Hosp Med (Lond)*. 2011;**72**(10):M146-51.
8. Gildeen DH. Infectious causes of multiple sclerosis. *Lancet Neurol*. 2005;**4**(3):195-202.
9. Heesen C, Mohr DC, Huitinga I, Bergh FT, Gaab J, Otte C, et al. Stress regulation in multiple sclerosis: current issues and concepts. *Mult Scler*. 2007;**13**(2):143-8.
10. Al-Mendalawi MD. A case of vocal tic: an unusual presentation of neurobrucellosis. *East Mediterr Health J*. 2013;**19**(10):898.