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Case Report

Neurobrucellosis Case Associated with Bilateral Abducens Nerve Involvement

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Abstract

Brucellosis is a zoonosis that causes chronic granulomatous infection. Human brucellosis is a systemic disease with a wide clinical spectrum. The clinical spectrum of the neurobrucellosis is very heterogeneous and neurologic manifestations can be seen in early or late disease. Central and peripheral nervous system involvement may also occur in 5-15% of patients. Due to the presence of different clinical manifestations and radiological findings, it can be confused with other neurological diseases. Neurobrucellosis case associated with bilateral VI. cranial nerve involvement which has not been previously mentioned in the literature was presented in this article.

ABBREVIATIONS

MRI: Magnetic Resonance Imaging; ESR: Erythrocyte Sedimentation Rate; CRP: C-Reactive Protein; CSF: Cerebrospinal Fluid; CNS: Central Nervous System; ADA: Adenosine Deaminase

INTRODUCTION

Neurobrucellosis can manifest in many different clinical forms. In the course of chronic brucellosis, while symptoms such as headache, fever, fatigue, tiredness are common, a direct neurological involvement is seen in about 5-15%. Acute, sub acute or chronic meningitis, meningoencephalitis, polyradiculoneuritis, myelitis and cranial nerve involvements are the most common neurobrucellosis clinics [1].

CASE PRESENTATION

Thirty-five years old male patient, engaged in farming, was admitted with complaints of double vision. Her complaints began 3 weeks ago. Accompanying joint pain, weakness and fatigue complaints were present. Brain MRI (Magnetic resonance imaging) was evaluated as normal in the first center where he applied upon beginning of his complaints and pain killers and vitamin supplements were given. Upon continuing complaints, any pathology was not detected except for mild anemia and high levels of erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP) in blood examinations performed in another center. Control brain MRI was normal again. Pyridostigmine and oral methylprednisolone treatments were started by thinking myasthenia gravis clinically. The patient whose complaints continued after ten days of treatment was admitted to our clinic. There was no known disease history of him but he had a birth story of the goats without using gloves three months ago. In the family history, it was learned that his brother was treated for brucellosis one year ago. Blood pressure was 130/80, fever was 36.5ºC and other examination findings were normal in his systemic examination. Any pathology was not detected in his neurological examination except finding of VI. Cranial nerve involvement (Figure 1-2). A mild anemia and slight high levels of sedimentation and CRP were observed in laboratory examination. In serum, brucellosis was (+) in tube agglutination test (Rose-Bengal) for titers of 1/160 and (+) in the Coombs test for titers of 1/320. In Cerebrospinal fluid (CSF) examination, CSF pressure was 160 mmH^2O, CSF direct smear was normal and glucose, chlorine and protein values were normal. CSF Brucella agglutination test was detected as 1/4 positive. PA lung graphy, ECG, echocardiography were normal. Brain MRI, chest CT, abdominal CT, which are radiological tests performed in the differential diagnosis, were normal. Repetitive nerve stimulation test performed with recording from the facial nerve innervated

Figure 1 Abduction limitation of right eye at right sight.

Figure 2 Abduction limitation of left eye at left sight.

orbicularis oculi and nasal cast in Electromyography was normal. Ceftriaxone, riphamycin and tetracycline were administered. Approximately one month later, the patient had a partial clinical improvement.

DISCUSSION

Brucellosis is an infectious disease that can transmit from animals to humans all over the world; particularly common in developing countries. Approximately 18,000 new cases of brucellosis have been reported in Turkey for every year. According to the geographical distribution, the prevalence of seropositive in Turkey ranges from 2.6% to 14.4% [2]. Brucella species are small gram-negative bacteria which can live and multiply inside mononuclear phagocytic system cells. This feature may explain long-term characteristic, complications and relapses of illness [3]. Brucellosis manifests itself with many different indications in acute and chronic periods. In acute period, fever, sweating, night sweats, back pain, headache, loss of appetite, joint pain, muscle pain, weakness, difficulty in walking, confusion, weight loss and numbness in the arm are observed [1]. In the base of chronic brucellosis, although symptoms such as headache, malaise, fatigue, depression are common, direct central nervous system (CNS) involvement is reported 5-15% in different series [1]. Brucella bacteria can hold the nervous system in direct manner or in an indirect way through cytokines and endotoxin [4]. Neurobrucellosis can be classified according to involvement of central nervous system, peripheral nervous system or both of them. Fever, vomiting, headache, neck stiffness may occur with or without systemic symptoms. Sensory and motor abnormalities in varying degrees, cranial nerve involvement, cerebellar dysfunction, coma and complications such as brain abscess can be seen. In neurobrucellosis course, although VIII. cranial nerve involvement is reported most frequently, II. VII. and VI. cranial nerve involvements are also reported [2,5]. In our case, bilateral VI.cranial nerve involvement was observed. In the literature, any case reporting bilateral cranial nerve involvement was not seen. When examining reasons for cranial nerve involvement in this case, etiology was determined with both high Brucella tube agglutination titer in blood and positive CSF Brucella tube agglutination titer together.

The fact that the clinical presentation cannot be explained by another disease, previous brucellosis history and the detection of antibodies against Brucella in blood and CSF with serum tube agglutination test and/or Coombs test are required for the diagnosis of neurobrucellosis. Lymphocytic pleocytosis and increased protein are present in the CSF and the growth in CSF cultures is positive in less than half of the cases [2]. Although adenosine deaminase (ADA) enzyme is not specific, it is detected high in CSF in neurobrucellosis cases. This enzyme can be detected as high in CSF in neurological involvements of lymphoma, tuberculosis, meningitis and sarcoidosis. In our case, brucellosis was (+) in tube agglutination test (Rose-Bengal) for titers of 1/160, (+) in the Coombs test for titers of 1/320 and CSF Brucella agglutination test was positive.

There is no full idea of unity between researchers for the treatment of neurobrucellosis. The drug selected should pass to the cerebrospinal fluid well and have bactericidal property. Although there is no full consensus, the administration of riphampycin or doxycycline together with dual or triple combinations of trimethoprim-sulfamethoxazole, ceftriaxone, streptomycin, gentamicin or ciprofloxacin antibioticsis recommended for 6-9 months [1]. In our case, ceftriaxone, riphampycin and tetracycline were administered and it was observed that the patient benefited from treatment in the control performed one month later.

Consequently, neurobrucellosisisanun common but serious complication of brucellosis. The variety of symptoms and signs can lead to delays in diagnosis and treatment. In our country where brucellosis is seen as endemic, neurobrucellosis should be kept in mind in the differential diagnosis of bilateral VI. cranial nerve involvement and similar clinical conditions. Because of the fact that there has not been any neurobrucellosis case presenting bilateral VI. cranial nerve involvement in the literature before, this case was found worthy to be presented.

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