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From a simple chronic headache to neurobrucellosis: a case report

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Abstract

Brucellosis is an infectious disease with high incidence in Iran. Neurobrucellosis is a focal complication of brucellosis affecting both central and peripheral nervous system presenting with a varieties of signs and symptoms. The most reported manifestations are meningitis and meningoencephalitis. In this report, we will describe a case of a young woman affected by neurobrucellosis presenting with chronic progressive headache and papilledema.

Keywords: Brucellosis, Headache, Papilledema.

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Introduction

Brucellosis is an infectious disease caused by the brucella bacteria. Brucella is an intracellular nonmotile gram-negative coccobacilli, which is able to have a prolonged intracellular life within phagocytes. Suppressed immunity leads to the organism proliferation and dissemination (1). Brucellosis is a zoonotic and occupationally related disease which can involve a variety of organs in human body such as gastrointestinal, genitourinary and central or peripheral nervous system. Human neurobrucellosis is now a rare disease in countries where eradication programs such as animal screening and vaccination are performed; but the high incidence rate of this disease in our country has made this area as an endemic region for brucellosis (2). This disease is mainly transmitted by dairy product such as raw milk, unpasteurized cheese and ice cream, direct contact to infected animals and inhalation of infected aerosolized particles (1-2). Neurobrucellosis is a focal complication of brucellosis which occurs in five to ten percent of cases with both central and peripheral nervous system involvement. Varieties of signs and symptoms have been listed for neurobrucellosis; but the most common manifestations are meningitis and meningoencephalitis**.** Neuropsychiatric syndromes have been report-

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ed in some rare conditions (3). Herein we will report a challenging case of neurobrucellosis presented by a chronic headache and papilledema.

Case report

A 25 year old woman was referred with a history of chronic headache for a period of three months. The headache was severe and refractory to NSAIDs or other pain relievers. There was no history of chills and fever, gait disturbance, hearing loss and memory problems. No history of using illicit drugs such as cocaine, alcohol, cannabis was also found. Drug history was negative for oral contraceptive pills, nalidoxic acid, tetracycline, corticosteroids, vitamin A and other drugs. She was student, lived in an urban area with history of travels to rural areas. No history of using unpasteurized milk and contact with animals were found.

In physical examination, vital signs were normal. Generally, she was cooperative with intact orientation. Cranial nerve examinations were normal and meningeal signs were not detected. There was a significant papilledema in ophthalmologic examination. Deep tendon reflexes (DTRs) in upper and lower limbs were normal. Muscle forces in proximal and distal regions of lower and upper limbs were also normal. No impairment in the sensation of extremities was detected. Gait was intact with no ataxia and tremor. Psychological examination was also done. There was not any abnormality in mood and affect evaluation and no cognitive impairment was also observed.

Laboratory studies including complete blood count with differentiation (CBC/diff), prothrombin time (PT), partial thromboplastin time (PTT), international normalized ratio (INR), blood urea nitrogen**,** creatinin**e** (BUN,Cr) and liver function tests (LFT) were normal. An elevated ESR was detected (68mm/hour). HBS Ag, HCV Ab, VDRL and HIV Ab were also negative. Anti-nuclear antibody (ANA), Anti-neutrophil cytoplasmic antibodies (ANCA), C3 and C4 were in normal range. Brain CT scan in addition to T1 and T2 weighted brain MRI plains after gadolinium injections were also performed that revealed no pathologic findings. The patient underwent lumbar puncture. Cerebrospinal fluid appeared xanthochromic and presented with a high opening pressure (280 mmH_2O), low glucose (43mg/dl) and elevated protein levels (338mg/dl). Spinal fluid analysis showed leukocytes. Brucella (capt test) tube agglutinin was positive at 1/2560 titers in blood and positive at spot and 1/2560 titers in CSF. Tuberculosis PCR test was negative for the CSF sample.

In summary, we had a case with a history of chronic headache characterized by increased intra-cranial pressure (ICP). Papilledema was the only positive finding in the initial physical examination. SAT (serum antibody test) was the only positive finding for brucella within the laboratory test. Other investigations including imaging studies were normal. The patient received intravenous cefteriaxone 1gr BID and oral doxycycline 100mg BID and rifampin 600 mg once daily for 1 month; and then rifampin continued for 14 months. Headache began to improve during treatment and disappeared after 3 months.

Discussion

Brucellosis considers as an important disease in endemic regions (4). Most commonly, ingestion of infectious products causes the disease but some patients are infected in occupationally related situations. Veterinarians, laboratory staffs, slaughter house workers and farmers are at a higher risk of this disease in comparison with normal population (5). Report of various and some rare symptoms and signs has made this condition as a challenging diagnostic problem. The diagnosis seems to be more problematic when the patients are presented with nonspecific symptoms such as fever, malaise and anorexia (6-7). Multiple organs can be involved in this condition including, gastrointestinal system (abdominal pain, gastroenteritis), blood (pancytopenia and coagulating disorders), pe-

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ripheral (PNS) and central (CNS) nervous system. Incidence of CNS involvement has been seen in 4-13% of patients. Meningitis, meningoencephalitis, polyradiculoneuritis and cranial nerve palsy are common symptoms; meningitis is the most common one. Stroke or hemorrhage can be occurred due to rupture of mycotic aneurisms caused by meningo-vascular involvement.

Guillian-barre syndrome (GBS), papilledema, increased intracranial pressure; cerebral venous thrombosis and sensory-neural hearing loss are other less common manifestations of CNS involvement. Additionally, white matter involvement is rare and can be in differential diagnosis of multiple sclerosis (MS) and progressive multifocal leukoencephalopathy (PML) (8-13).

The most practical way to diagnose neurobrucellosis is to keep this disease in mind especially in endemic regions and it should be considered in patient with chronic slowly progressive neurologic sign and symptoms. Exact diagnosis of neurobrucellosis depends on abnormal CSF and blood findings indicating: a) CSF lymphocytic pleocytosis and increased protein, b) positive CSF culture for the Brucella organisms, c) positive Brucella IgG agglutination titer in the blood and d) response to specific chemotherapy with a significant drop in the CSF lymphocyte count and protein concentration. The most sensitive way to confirm the diagnosis is to detect antibodies against brucella bacteria in serum or CSF fluid (14-15).

Neurobrucellosis may be presented without any specific systemic symptoms or laboratory findings in some cases. Positive blood and CSF cultures can prove the diagnosis but with a low sensitivity especially in chronic brucellosis. On the other hand, other cultures need a longer time to detect the organisms; therefore, CSF and blood cultures can be negative. Considering above, the diagnosis is generally based on serological methods. Presence of the brucella antibody in the CSF has a high diagnostic value (14). Moreover, clinical improvement following antibiotic therapy can be a good support to prove the diagnosis (15). Radiologic studies are not too sensitive. They are normal in many cases. Inflammation and abnormal enhancement of the white matter and vascular changes are other uncommon results (16). In our case, imaging studies were normal. We detected only a chronic headache in history and papilledema in physical exam; but serum antibody test (SAT) was positive in first step.

In conclusion, brucellosis is still endemic in some countries and physicians in these regions must consider that this infection may involve any organ and may vary markedly in its clinical presentation. Neurobrucellosis should be considered in chronic progressive neurological symptoms such as headache, cognitive dysfunction, paraparesis, and transient ischemic attacks. In endemic regions patients who presented by pure neurological signs or symptoms should be assessed for neurobrucellosis as early diagnosis and treatments lead to a decreased complications.

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