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Case Report / Olgu Sunumu

A Case of Neurobrucellosis That Mimicks Increased Intracranial Hypertension Syndrome

Kafa İçi Basınç Artma Sendromunu Taklit Eden bir Nörobruselloz Olgusu

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Summary

Neurobrucellosis can present with various and misleading clinical symptoms and signs. We present a female patient who was hospitalized because of headache that was different from her previously migraine headaches. The patient had epileptic seziures, diplopia, bilateral papilledema that benifited from treatment. (Turkish Journal of Neurology 2014; 20:132-134)

Key Words: Neurobrucellosis, intracranial hypertension syndrome, diplopia, headache, papiledema

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Özet

Nörobruselloz olguları farklı ve yanıltıcı klinik semptom ve bulgular sergilemektedir. Biz aşağıda önceden migreni olup migrenden farklı bir baş ağrısı, epileptik nöbet, diplopi ve papilödem nedeniyle yatırılan, etiyolojisinde bruselloz saptanan ve tedaviden yarar gören bir olguyu sunuyoruz. (Türk Nöroloji Dergisi 2014; 20:132-134)

Anahtar Kelimeler: Nörobruselozis, intrakraniyal hipertansiyon sendromu, diplopi, başağrısı, papilödem

Çıkar çatışması: Yazarlar bu makale ile ilgili olarak herhangi bir çıkar çatışması bildirmemişlerdir.

Introduction

Brucellosis is a zoonosis that is transmitted from animals to humans through a bacteria called brucella. The disease can be transmitted by consuming the meat, milk or milk products produced from an infected animal, or by contact with placenta of the animal. Systemic symptoms emerging either acutely or after 2-4 weeks incubation period include shivering and fever, muscle and large joint pains, sweating and fatigue (1).

Clinical manifestations of brucellosis vary significantly. The cases with nervous system involvement are called neurobrucellosis (NB). Neurobrucellosis can occur in 1/3rd of the cases either at the onset or at any point throughout the course of the disease. Neurobrucellosis can cause convulsions, paralysis, ataxia, hearing impairment, vertigo, headache, meningoencephalitis, myeloradiculoneuritis, brain abscess, epidural abscess, papillary edema, hydrocephalus, pseudotumor cerebri, orientation difficulty and personality changes (2,3,4,5,6). The most common manifestation of NB is meningitis (7).

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Case

Fourty two years old female patient who had a history of migraines came to our emergency room with generalized tonic-clonic seizure and 30-minute bout of loss of consciousness. After being monitored for a while in the emergency room, she was discharged but she returned with adductive deviation of her left eye, for which she was seen by a neurologist and optometrist at the clinic. The deviation disappeared after one and a half months. She then came back with blurry vision and was diagnosed with bilateral papilledema. After she was referred to the neurology policlinic, she was admitted with the prediagnosis of increased intracranial pressure (IIP). Her anamnesis indicated that she consumed cheese produced from unpasteurized milk. She also received total thyroidectomy six years ago and has been continuing B12 replacement therapy. She was also using ergot for migraine. She did not have a history of feverish diseases or epileptic seizures. There were no meninx irritation findings in her neurological exam. Bilateral papilledema (Figure 1) and irregular visual field defects were present. Blood biochemistry and hemogram were within normal ranges. Because of the IIP prediagnosis, she went under magnetic resonance (MR) imaging and MR venography which were both interpreted as normal. Lumbar puncture was made due to the lack of any extraordinary mass and the normal appearance of the ventricles. Opening pressure of cerebrospinal fluid (CSF) was 170 mmH2O and within normal ranges. In CSF, protein was 176 mg/dl, glucose was 26 mg/dl (simultaneous blood glucose 112 mg/dl), chlorine was 120 mmol/l and sodium was 143 mg/dl. There were no cells in direct CSF microscopy. Brucella spp reproduced in the CSF culture. In addition, blood culture, VDRL, Hepatitis A, B, C, HIV serology, and antinuclear antibody (ANA) were found to be negative. Brucella tube agglutination test was 1/80. Papilledema indicators such as vitamin A deficiency or excess or iron deficiency anema have not been found. Throid function tests were within normal ranges. She had not been using drugs such as lithium, tetracycline or corticosteroids. The patient who was diagnosed with brucellosis on the basis of laboratory findings was evaluated as NB due to non-migraine headache, epileptic seizure, recovered possible left abducens nerve paralysis and papilledema findings. She was started on a triple antibrucellosis treatment consisting of rifampicin, doxycycline and ceftriaxone. After the treatment, headache, nausea, vomiting diseappeared completely. There were partial improvements in papilledema and visual area defects (Figure 2).

Figure 1. Papilledema seen in the first retinal imaging

Figure 2. Reduced papilledema in the six-month follow-up retinal image

Discussion

First, we thought we had a case of IIP with headache, epileptic seizure, abducens paralysis and papilledema. After normal MR and CSF opening pressure and laboratory studies, however, we diagnosed our case as NB because of the production of Brucella spp in CSF.

Neurobrucellosis can occur with neurological findings in the absence of systemic symptoms and findings (8). Central nervous system involvement in brucellosis, known as NB, is a rare complication with 3-5% incidence rate. Neurobrucellosis has a wide range of clinical manifestations including meningitis, meningoencephalitis, myelitis, epidural abscess, spinal abscess, optical neuropathy, peripheral neuropathy, depression and psychosis (9). The frequency of NB among systemic brucellosis cases was reported to be e-25% (1). Neurological symptoms can sometimes be the only symtoms of brucellosis. Due to NB, the patients may present with convulsion, paralysis, ataxia, hearing deficit, vertigo, headache, orientation impairment and personality changes (9). The fact that migraine headache in our case was replaced by a different type of headache and the tonic-clonic seizure were clinically evaluated as NB symptoms.

Brucellosis diagnosis is made on the basis of epidemiological and clinical findings as well as bacteriological and serological test results. The symptoms of the disease can mimic many others and misdiagnoses are possible due to its complications. Diagnosis of NB is made after growing the bacteria in CSF and/or standard tube agglutination test. Cerebrospinal fluid findings in NB are primarily in the form of pleocytosis dominated by lymphocytes, low glucose levels and high protein levels (5,7). Lymphocytic pleocytosis in varying amounts in CSF are abnormal for almost all of NB cases. We did not detect yymphocytic pleocytosis in the CSF study of our case. There are limited number of reported instances of this in the literature (10).

Serum and CSF antibodies against brucella antigens can be detected in most of the cases (11). Even though general consensus suggests positive cell culture as the gold standard for diagnosis, it is often thought to be suboptimal. Gül et al. found positive blood cultures for 28% of the patients and positive CSF cultures for 14% of the patients (12). Similarly, another study found positive CSF culture in less than 15% of NB cases (13). The diagnosis was made upon the growth of the bacteria in the CSF of our patient who had scattered nervous system findings.

Meningitis is the most common clinical presentation of NB. Yılmaz et al. reported central nervous system involvement in only 10 out of 140 brucellosis patients over the course of 10

years (1.48%). Recurring meningitis syndrome in addition to epileptic seizures were reported in one of those cases (14). Gül et al. reported convulsions in 8% of 187 NB cases (11). In our case, as well, there was a one-time generalized tonic-clonic epileptic seizure incident. Antiepileptic treatment was not considered for our case whose seizure was evaluated as symptomatic and non-recurring.

Through the course of the disease, our case possibly had diplopia due to left nervus abducens paralysis prior to the treatment. Cranial nerve involvement is a rare complication in NB. The most commonly involved cranial nerves are optical, oculomotor, facial, abducens and vestibulocochlear nerves. Sensoryneural hearing loss, facial paralysis and diplopa can be seen (15). Gül et al. reported cranial nerve involvement in 19% of the patient among with 14 had 6th cranial nerve involvement (11). Espejo et al. reported nervus abducens paralysis as a clinical manifestation of NB (16).

Even though our case had IIP findings, CSF opening pressure was measured to be 170 mmH2O. Some cases may in fact show normal CSF opening pressure just like our case who also had pseudotumor cerebri findings (6,19). Pressure of CSF may fluctuate and lumbar puncture should be repeated if there is continuing clinical suspect (16). Lumbar puncture was not repeated in our patient due to her refusal of the procedure. She was followed on the basis of clinical parameters.

Papilledema is among the clinical presentations of NB. Gül et al. reported papilledema in 3% of their cases (11). The appearance of papilledema can remain unchanged for months or even years (17). McLean et al. detected papilledema in 6 out of 18 NB cases and reported that this finding lasts somewhere between one week and 8 months (18). In the follow-up visits after triple antibiotherapy, it was seen that the complaints were mostly resolved and papilledema still kept improving after six months.

In Turkey where brucellosis is endemic, NB should be included in the differential diagnosis for the cases that present with central nervous system findings but with normal imaging studies. The presence of clues indicative of NB in the patient history should facilitate easy diagnosis.

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