Bilateral papilledema caused by *brucellosis* mimicking pseudotumor cerebri

Psödotümör serebriyi taklit eden brusellozun neden olduğu bilateral papil ödem

Tuğçe MENGI, Mehmet ÇELEBİSOY

Summary

In this article, we report a patient with migraine who was hospitalized with a prediagnosis of pseudotumor cerebri and diagnosed as neurobrucellosis with isolated intracranial hypertension presentation. A 22-year-old woman was admitted to emergency department with a complaint of headache. Her anamnesis indicated that she had migraine for seven years. Neurological examination revealed bilateral papilledema. Cranial magnetic resonance imaging was normal. Cerebrospinal fluid examination revealed 80 lymphocytes per mm³ with 178 mg/dL protein. Opening pressure was 260 mmH₂O. *Brucella* tube agglutination and Rose Bengal tests were positive in blood and cerebrospinal fluid. She was diagnosed as neurobrucellosis. If the systemic findings are insignificant and neurological findings are atypical such as isolated papillary edema, neurobrucellosis may not be considered and its diagnosis may be delayed. We believe that *brucella* serology should be included in the diagnostic protocols in endemic areas. Thus, early diagnosis and appropriate treatment can prevent complications of neurobrucellosis.

Keywords: Brucellosis; headache; meningitis; migraine; pseudotumor cerebri; papilledema

Introduction

Idiopathic intracranial hypertension is characterized by elevated intracranial pressure of unknown etiology with normal cerebrospinal fluid (CSF) composition.[1] Pseudotumor cerebri includes secondary causes of intracranial hypertension in the absence of hydrocephalus, mass, or structural lesion and no abnormal meningeal enhancement on magnetic resonance imaging (MRI).[1,2] Idiopathic intracranial hypertension is a subset within the primary pseudotumor cerebri category, while the secondary pseudotumor cerebri group would include cause such as medical conditions, medications and exposures, cerebral venous abnormalities (e.g. decreased CSF absorption from previous intracranial infection or subarachnoid hemorrhage).[2]

Chronic migraine is characterized by recurrent headache attacks that happen 15 or more days per month, 8 of which with migraine features.[3] Migraine mimics includes two different groups. First group is primary headache conditions such as the trigeminal autonomic cephalgias, while second group is important to differentiate because of their underlying causes such as intracranial mass lesions, raised intracranial pressure, and meningitis.[4]
In this article, we report a patient with migraine who was hospitalized with a prediagnosis of pseudotumor cerebri and diagnosed as neurobrucellosis with isolated intracranial hypertension presentation.

Case Report

A 22-year-old woman was admitted to our emergency department with a complaint of headache. Her anamnesis indicated that she had migraine without aura for seven years, headaches aggravated with vomiting in the last month. She was admitted in the separate institutes due to continuous headache and was diagnosed with chronic migraine.

Physical examination did not reveal any pathology such as lymphadenomegaly or organomegaly. Neurological examination was normal except bilateral papilledema. There was no fever, neck stiffness, Kernig’s sign, change in mental status. Cranial computed tomography revealed no pathology. She was hospitalized with the prediagnosis of pseudotumor cerebri. Her anamnesis indicated that she consumed cheese produced from unpasteurized milk. Blood tests included blood count, renal and liver function were within normal ranges. Erythrocyte sedimentation rate was 6 mm/h, and C-reactive protein was negative. Cranial MRI and MR venography showed no pathology. Bilateral enlarged blind spot was detected in the visual field. The visual acuity was 20/20. Lumbar puncture was performed and opening pressure was 260 mmH₂O. CSF examination revealed 80 lymphocytes per mm³, with 178 mg/dL protein and 16 mg/dL glucose (serum glucose: 101 mg/dL). Brucella tube agglutination test was positive at 1/320 titers in blood and positive at 1/40 titers in CSF. Serum and CSF Rose Bengal tests yielded positive results. There was no growth in CSF culture. She was diagnosed as neurobrucellosis and was treated with ceftriaxone (2 g/day), rifampicin (600 mg/day) and doxycycline (200 mg/day). Chest X-ray, abdominal ultrasonography and lumbosacral MRI were performed for subclinical involvement, and they were unremarkable. Her headache was improved in the follow-up period. In the control examination after three months, the optic discs were normal.

Discussion

Brucellosis is a multisystemic disease that primarily affects the musculoskeletal, hematopoietic, genitourinary, cardiovascular, respiratory, and nervous systems. Neurobrucellosis generally occurs as the addition of neurological complications on existing systemic symptoms.[9] The patients with isolated neurological symptom are usually limited to case reports.[5–9]

Different classifications were used evaluating neurobrucellosis.[10] In a classical review on this subject, Gul et al.[11] divided major complications of neurobrucellosis into cranial nerve involvement, polynuropathy/radiculopathy, depression, paraplegia, stroke, and abscess formation. The report by Demiroğlu et al.[10] classified into four groups as meningitis, encephalomyelitis, polyradicular group, and others. The classification is difficult because of brucellosis may mimic a large number of central and peripheral nervous system pathologies.

The clinic of brucella meningitis is generally characterized by fever, headache, and nuchal rigidity.[5] CSF biochemistry and cytology, CSF culture, serological tests in CSF can be diagnosed.[10] Although demonstration of brucella from CSF culture is the gold standard for diagnosis, positive test is detected in few cases.[11,12] Lymphocytic pleocytosis, decreased glucose level, and elevated protein content are seen in the CSF.[9,10,13] Presumptive diagnosis can be made serologically by Rose Bengal and standard tube agglutination tests.[6]

Papilledema occurs in 3% of cases of neurobrucellosis.[11] Intracranial hypertension or optic neuritis have been involved in papilledema pathophysiology.[7,11] The initial clinical presentation of neurobrucellosis with intracranial hypertension rarely has been reported.[5–7,13–18] In a study that includes 187 cases of neurobrucellosis, intracranial hypertension were 0.5%. Güngör et al.[19] reported the association between pseudotumor cerebri and brucellosis. Papilledema and the other findings of increased intracranial pressure may develop due to meningitis or meningencephalitis.[5,7,14,15,20,21] There was no eye pain in our case. There were not a decrease in visual acuity. The pupils were reactive with no afferent defect. Opening pressure was 260 mmH₂O. Our case had with the typical signs and symptoms of intracranial hypertension and atypical clinical findings for meningitis. There was no fever, neck stiffness, Kernig’s sign or change in mental status. She had abnormal CSF composition.
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but fulfilled all the other diagnostic criteria for pseudotumor cerebri syndrome so that the cause of intracranial hypertension was investigated. *Brucella* tube agglutination test was positive at 1/320 titers in blood and positive at 1/40 titers in CSF. Serum and CSF Rose Bengal tests yielded positive results. Our case with findings of isolated intracranial hypertension was diagnosed as neurobrucellosis.

Treatment of neurobrucellosis includes more than one antibiotic. Doxycycline, rifampicin, ceftriaxone and co-trimoxazole can cross the blood-brain barrier. Thus a good CSF concentration can be achieved. The concentration of streptomycin/gentamycin in CSF is therapeutic only when meninges are inflamed. The generally approved protocol for treatment is the administration of doxycycline in combination with two or three drugs, e.g. rifampicin, co-trimoxazole, ceftriaxone, that can cross the blood-brain barrier for many months (3–12 months). The prognosis is usually good with an improvement of symptoms if appropriate treatment is started early. Sequelae occur mostly in patients where treatment had been started late.

In conclusion, if the systemic findings are insignificant and neurological findings are atypical such as isolated papillary edema, neurobrucellosis may not be considered and its diagnosis may be delayed. We believe that *brucella* serology should be included in the diagnostic protocols in endemic areas like Turkey. Thus, early diagnosis and appropriate treatment can prevent complications of neurobrucellosis.

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