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CASE REPORT

Bilateral papilledema caused by *brucellosis* mimicking pseudotumor cerebri

Psödotümör serebriyi taklit eden brusellozun neden olduğu bilateral papil ödem

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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 7 years. Neurological examination revealed bilateral papilledema. Cranial magnetic resonance imaging was normal. Cerebrospinal fluid (CSF) examination revealed 80 lymphocytes per mm^3 with 178 mg/dL protein. Opening pressure was 260 mmH_2O. *Brucella* tube agglutination and Rose Bengal tests were positive in blood and CSF. 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.

Özet

Bu yazıda, psödotümör serebri ön tanısıyla hastaneye yatırılan ve izole intrakraniyal hipertansiyon prezentasyonu ile nörobruselloz tanısı alan migrenli bir hastayı sunuyoruz. Yirmi iki yaşında kadın hasta, acil servise baş ağrısı yakınmasıyla başvurdu. Öyküsünde yedi yıldır migren tanısı olduğu belirtildi. Nörolojik muayenesinde bilateral papilödem saptandı. Kraniyal manyetik rezonans görüntüleme normaldi. Beyin omurilik sıvısı incelemesinde mm^3 başına 80 lenfosit ile 178 mg/dL protein saptandı. Açılış basıncı 260 mmH_2O idi. *Brucella* standart tüp aglütinasyonu ve Rose Bengal testleri kanda ve beyin omurilik sıvısında pozitifti. Nörobruselloz tanısı koyuldu. Sistemik bulguların silik, nörolojik bulguların izole papilödemi gibi atipik olması durumunda nörobruselloz düşünülmeyebilir ve tanısı gecikebilir. Endemik alanlarda tanı protokolleri içinde *Brucella* serolojisinin yer alması gerektiğini düşünüyoruz. Böylece, erken tanı ve uygun tedavi ile nörobruselloz komplikasyonlarının önüne geçilebilir.

Anahtar sözcükler: Bruselloz; baş ağrısı; menenjit; migren; psödotümör serebri; papilödem.

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, and cerebral venous abnormalities (e.g., decreased CSF absorption from the 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

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include 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]

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 7 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, and 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 mmH_2O. CSF examination revealed 80 lymphocytes per mm^3, 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 3 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.[5] 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, polyneuropathy/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 has 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 was 0.5%.[11] 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 meningoencephalitis.[6,7,14,15,20,21] There was no eye pain in our case. There was not a decrease in visual acuity. The pupils were reactive with no afferent defect. Opening pressure was 260 mm H_2O. 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 but fulfilled all the other diagnostic criteria for pseudotumor cerebri syndrome [22] 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.[10,20] The concentration of streptomycin/gentamycin in CSF is therapeutic only when meninges are inflamed.[20] The generally approved protocol for treatment is the administration of doxycycline in combination with two or three drugs, for example, rifampicin, co-trimoxazole, and ceftriaxone that can cross the blood-brain barrier for many months (3–12 months).[10] The prognosis is usually good with an improvement of symptoms if appropriate treatment is started early.[8,20] Sequelae occur mostly in patients where treatment had been started late.[8]

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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