Pseudotumor cerebri due to brucellosis: A rare case report

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Abstract

Neurobrucellosis is a focal form of brucellosis that may be life-threatening if not timely diagnosed and treated. Pseudotumor cerebri (PTC) has rarely been reported as the sole manifestation of brucellosis. A 41-year-old woman came to the hospital with chronic headaches, vertigo, and nausea. She had been experiencing symptoms for 8 months, starting with blurred vision and headache. The diagnosis was pseudotumor cerebri, managed with acetazolamide and topiramate. She had bilateral papilledema but a normal chest and cranial nerves exam. Lab tests were normal except for high ESR. Brain scans showed no issues. A lumbar puncture was done to reduce intracranial pressure. High ICP made CSF shunt necessary to prevent visual loss. However, a serum brucella agglutination test was done due to chronic neurological symptoms and lymphocyte dominance in CSF analysis. A positive result with the Wright test and 2ME titer. After two weeks, the patient was treated with gentamycin, ceftriaxone, doxycycline, and rifampin, and improved without neurosurgical intervention. Here, we have reported a case of brucellosis with severe headaches, progressive visual impairment, and fundoscopic finding of papilledema, later diagnosed as brucellosis-induced PTC.

Keywords: Neurobrucellosis; Pseudotumor Cerebri; Intracranial pressure

Key Clinical Message

Severe headaches, progressive visual impairment, and fundoscopic findings of papilledema, later diagnosed as pseudotumor cerebri, could be the sole manifestations of brucellosis.

Introduction

Brucellosis is a zoonotic infectious disease with various manifestations. This infection is endemic in the Middle East, especially Iran. Brucellosis can be treated readily. However, relapses and chronicity are prevalent (1). Neurobrucellosis is a focal form of brucellosis that may be life-threatening if not timely diagnosed and treated. This complication of brucellosis can present with various symptoms such as headache, meningismus, nausea and vomiting, diplopia, lethargy, and even coma. Meningoencephalitis is the most prevalent feature of neurobrucellosis. Nonetheless, cranial nerve palsies, brain abscess, cerebral venous thrombosis, subdural hemorrhage, Guillain-Barre syndrome, diabetes insipidus, myelitis, radiculoneuritis, and peripheral neuropathy are some other uncommon syndromes (2-5). Pseudotumor cerebri (PTC) has rarely been reported as the sole manifestation of brucellosis (6). Here, we have reported a case of brucellosis with severe headaches, progressive visual impairment, and fundoscopic finding of papilledema, later diagnosed as brucellosis-induced PTC.

Case presentation

A 41-year-old woman presented to the hospital with chronic headaches, vertigo, and nausea. Her symptoms had started 8 months earlier with blurred vision and headache, which was later diagnosed as pseudotumor cerebri and was controlled with acetazolamide and topiramate. Her vision acuity further decreased in the past 2 months ago, and myalgia and intermittent fever were added to the previous complaints. She was conscious at presentation with no focal neurological deficit. However, her physical examination revealed bilateral papilledema. The chest test and cranial nerves examination were all normal. At presentation, routine laboratory tests were all in the normal range except for an increased erythrocyte sedimentation rate (ESR) (45 mm/h) and lactate dehydrogenase (LDH) (540 U/ml). Magnetic resonance imaging and computed tomography of the brain revealed no occupying lesion or other abnormalities. A lumbar puncture was performed in order to decrease the intracranial pressure. The cerebrospinal fluid (CSF) examination revealed an opening pressure of more than 40 cmH2O, white blood cell (WBC) count of 87 cells/mm (79% lymphocytes and 21% neutrophils), and protein and glucose concentrations of 43 and 83 mg/dl, respectively. Due to this significantly increased ICP, she was a candidate for CSF shunt to prevent further visual loss. Nevertheless, considering the patient's chronic neurologic symptoms and also the lymphocyte dominancy of the CSF analysis, a serum brucella agglutination test (BAT) was done, which demonstrated a positive Wright test with a titer of 1:80 and 2ME titer of 1:40. Interestingly, the CSF wright test was also positive with a titer of 1:40. Hence, she was started on treatment with gentamycin, ceftriaxone, doxycycline and rifampin with the diagnosis of neurobrucellosis. She gradually improved clinically after two weeks without needing CSF shunting or other neurosurgical intervention.

Discussion

Brucella species are intracellular pathogens causing systemic infection in humans and livestock. Brucellosis can involve the central nervous system through direct damage or indirect induction of immune neuroinflammation (7). Neurobrucellosis usually presents with classic meningoencephalitis syndrome. However, we should consider the diagnosis in any patient presenting with chronic headache or neuropsychiatric symptoms, even without signs of meningeal irritation or fever (8). Neurobrucellosis is also suspected in patients with chronic neurological symptoms accompanied by CSF lymphocytosis or compatible neuroimaging findings. Nevertheless, it is confirmed by positive serum brucella agglutination test, positive serological tests (increased brucella antibody in the CSF), positive CSF Wright test, and isolation of Brucella species or detection of brucella DNA in the CSF with polymerase chain reaction (PCR) test (9). Our patient had been previously diagnosed with idiopathic PTC. Nonetheless, her aggravating symptoms and the addition of systemic manifestations like fever and myalgia suspected us of the diagnosis of brucellosis. In addition, the CSF lymphocytosis made our suspicion to neurobrucellosis stronger.

PTC has been a rare presentation of brucellosis. PTC is characterized by elevated ICP with normal CSF cell count, biochemistries, and normal brain imaging findings. It has been more commonly reported in females of childbearing age and obese individuals(10, 11). PTC may be primary or secondary to traumatic brain

injuries, collagen vascular disorders like systemic lupus erythematosus (SLE), medications like tetracyclines, isotretinoin, phenytoin, and other medical conditions (12-15). Our patient was initially diagnosed as being involved with primary or idiopathic PTC. However, brucellosis was later identified as the causative condition for her problem. Previously, rarely reported infections as the underlying causes of PTC included measles, varicella, Lyme disease, human immunodeficiency virus (HIV), and tuberculosis (16-21). However, brucellosis has been more commonly reported recently as the trigger for PTC, compared with other mentioned infections (6, 22-26). As aforementioned, neuroimaging usually offers normal findings in PTC.

Nonetheless, flattening of the globe, empty sella, enlarged prelaminar optic nerve or optic nerve sheath (ONS), enhanced optic nerve head, increased tortuosity of the optic nerve, distension of the perioptic subarachnoid space, narrowing of Meckel's cave and sinovenous stenosis may be identified as indicators of PTC (27-29). Treatment of PTC is based on controlling the underlying condition, medical management, repeated lumbar punctures, and surgical interventions(11). However, PTC in the settings of brucellosis can readily be controlled by antibiotic therapy with favorable results if timely diagnosed and not led to sequels.

On the other hand, delayed diagnosis and management of PTC may result in poor visual outcomes (30). The best therapeutic regimen has not been determined for neurobrucellosis. Nevertheless, at least a dual- or triple therapy with a combination of streptomycin, ceftriaxone, doxycycline, rifampicin, and trimethoprim-sulfamethoxazole for at least 6 months is recommended. Ceftriaxone-based therapeutic regimens have yielded the highest cure rates for neurobrucellosis(31). Our patient was initially responsive to CSF volume-decreasing pharmaceuticals but was later found to require surgical management. Nevertheless, she was promptly started on combination antimicrobial therapy after brucellosis was recognized as the underlying cause of her condition. Fortunately, she responded favorably to medical treatment and was deprived of the need to undergo surgical shunt implantation.

Conflict of Interest

None.

Acknowledgments

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Written informed consent statement

Written informed consent was obtained from the patient for publication of the current case report.

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