Journal of Case Reports in Practice (JCRP) 2017; 5(4): 33-35

CASE REPORT

Neurobrucellosis: a case report of papillophlebitis

Nafiseh Mohebi[1], Mehdi Moghaddasi[1], Peyman Daraee[1]

[1]*Department of Neurology, Rasoul-Akram Hospital, Iran University of Medical Science (IUMS), Tehran, Iran*

ABSTRACT

Ocular involvement is an infrequent presenting feature in brucellosis; consequently, the exact incidence has not been established. Herein, we report a 29-year-old woman diagnosed with neurobrucellosis presenting with aseptic meningitis and unilateral blurred vision in whom simultaneous asymptomatic papillophlebitis and symptomatic optic atrophy had been established. To the best of our knowledge, this is the first reported case of neurobucellosis associated with papillophlebitis. It is worth to mention that discovering neurobrucellosis especially while manifesting with rare features, requires high suspicious of diagnosis peculiarly in endemic countries as Iran. In our case, positive serum and cerebrospinal fluid (CSF) Wright, Coombs Wright and 2-Mercapto Ethanol (2ME) confirmed neurobrucellosis.

Key words: papillophlebitis, neurobrucellosis, chronic meningitis, brain white matter lesion

INTRODCTION

Brucellosis is a zoonotic infection caused by bacterial genus Brucella. The bacteria are transmitted from animals to humans by ingestion through infected food including unpasteurized milk products, undercooked infected meats and direct contact with an infected animal or inhalation of aerosols.[1] Brucellosis is the most common bacterial zoonosis with widespread geographic distribution and is labelled as regionally emerging zoonotic disease. The high prevalence in certain geographic areas is well recognized, especially in Mediterranean countries and the Middle East, including Iran.[2] Neurobrucellosis is the complication of a systemic infection. The frequency of neurobrucellosis has been reported as 5–7% in literature.[1] Neurological complications include encephalitis, meningoencephalitis, radiculitis, myelitis, peripheral and cranial neuropathies, subarachnoid hemorrhage, psychiatric manifestations, brain abscess, and demyelinating syndrome.[3] Ocular, optic nerve and chiasmatic involvement are extremely rare. Reported here is a rare case of neurobrucellosis in a young woman manifesting with visual impairment diagnosed with papillophlebitis.

CASE REPORT

A 29-year-old woman, living in a rural area of Iran,

Correspondence:
Nafiseh Mohebi
Department of neurology, Rasoul-Akram Hospital, Iran University of Medical Sciences, Tehran, Iran
Tel: +98 912 2898013
Email: nfs.mohebi@gmail.com

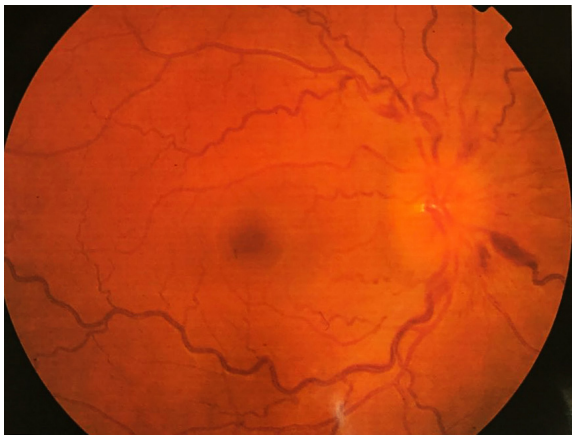

Figure 1, Right optic edema associated with splinter hemorrhage and vasodilation of retinal veins.

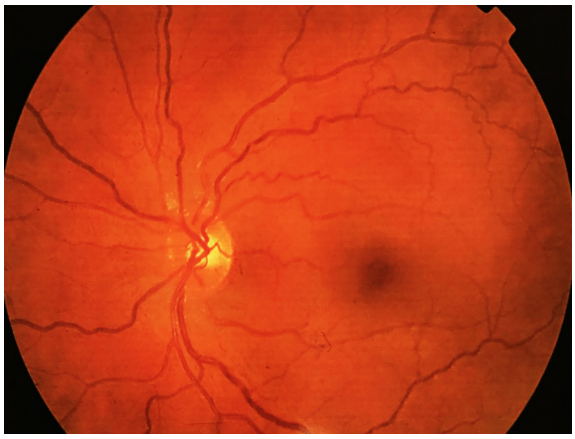

Figure 2, left optic disc with mild pallor compatible with optic neuropathy.

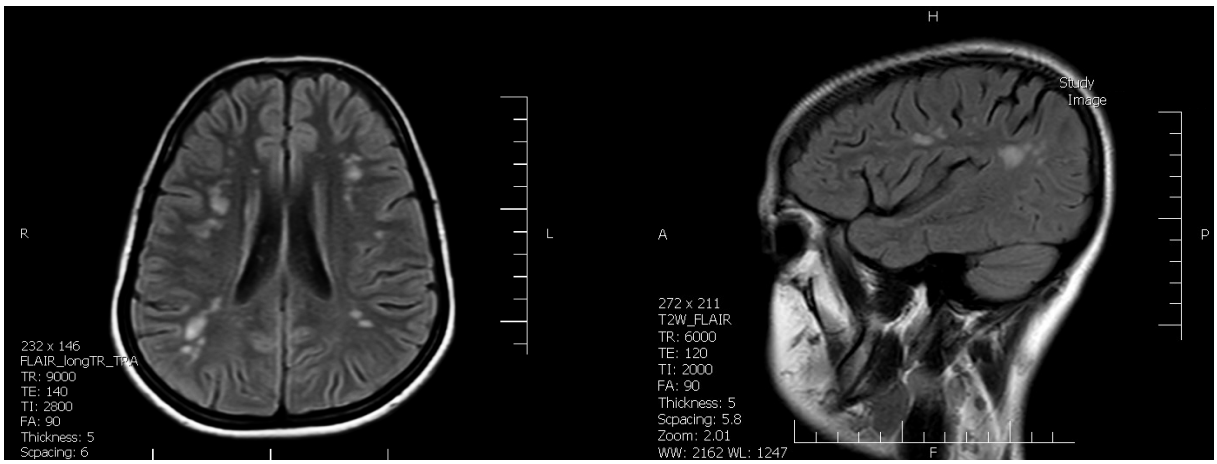

Figure 3, Brain MRI (FLAIR) showing bilateral subcortical white matter demyelinating lesions (Left: axial, Right: sagittal).

presented with painless and acute visual loss of left eye since one month earlier, predominantly involving peripheral visual field. She did not have any complain in right eye. Her past medical history was significant for migraine type headache since two years ago for which amitriptyline was prescribed. Recently, she had been experiencing more severe headaches with no relief with medication. The patient was afebrile without organomegaly or arteritis. Mental status examination showed mild depression with intact cognition. Visual acuity was 8/10 and 3/10 in right and left eye respectively accompanied with diminished color vision bilaterally. Ophthalmoscopic examination of right eye, the asymptomatic one, revealed optic disc edema with splinter hemorrhage and vasodilation of retinal veins so, papillophlebitis was diagnosed (Fig. 1). The left optic disc, the symptomatic eye, showed mild disc pallor compatible with optic neuropathy (Fig. 2). External ocular movements, other cranial nerves, motor forces, sensory, cutaneous and deep tendon reflexes, skin and joints were entirely normal. Lumbar punctuation (LP) was performed with opening pressure of 23CmH2O and cerebrospinal fluid (CSF) analysis revealed pleocytosis (120/cm^2) with lymphocytes predominance, increased protein (110 mg/dL) and decreased glucose levels (46 mg/dL). CSF culture was negative. Brain magnetic resonance imaging (MRI) showed symmetric subcortical hypersignal lesions in T2/FLAIR (Fig. 3). Anterior visual pathway, brain magnetic resonance angiography (MRA), cervical MRI, chest X-ray and transthoracic echocardiography were all unremarkable. Regarding CSF composition and history of unpasteurized dairy product consumption, neurobrucellosis was suspicious. Results of serologic test including serum Coombs Wright (1:320) and 2-ME IgG (1:640) and CSF Wright (1:80), Coombs Wright (1:160) and 2-ME IgG (1:80), all are supposed to be positive in an endemic country as Iran. Other infectious screening tests including PPD test, VDRL, HSV, VZV, CMV and EBV PCRs, hepatitis viruses and vasculitic tests were all negative. The patient was diagnosed with neurobrucellosis and antibiotics including ceftriaxone, rifampin and doxycycline had been commenced for two weeks accompanied with dexamethasone folowed by doxycycline,trimethoprim-sulfamethoxazole and rifampin. One week after treatment visual acuity and headache had been obviously improved with minor amelioration in ophthalmoscopic examination.

DISCUSSION

Brucellosis is a common zoonosis that can infect humans if unpasteurized dairy products are consumed, especially milk and its products.[1] It is still endemic in many parts of the world including coastal countries of Mediterranean Sea, Middle East regions and Central and South Americae,[1,4] there is no typical clinical picture and specific CSF findings for neurobrucellosis. The best diagnostic methods are blood and bone marrow cultures.[1,5] Considering the history of unpasteurized milk or milk products ingestion and lack of typical signs of a known neurological disease, neurobrucellosis is worth to be ruled out. Serum agglutination titer greater than 1:160, CSF agglutination titer greater than 1:80, CSF composition compatible with aseptic meningitis, all can establish the diagnosis.[3,6] However, in endemic areas, serum agglutination titer greater than 1:80 can be considered positive6 and Iranian expert consensus advocates that any positive CSF agglutinin titer is diagnostic for brucellosis. Clinical categories of neurobrucellosis are variable including acute, subacute or chronic meningitis, meningoencephalitis, arachnoiditis, myelitis, polyradiculitis, mononeuritis and vasculitis. Documented cranial nerve involvement includes vestibulocochlear, trigeminal, facial, abducens, oculomotor and optic nerves, among which optic nerve involvement is the most uncommon.[3] Ocular involvement in brucellosis is variable and include dacryoadenitis, conjunctivitis, episcleritis, keratitis, neuroretinitis, choroiditis, panuveitis, pars planitis and hyalitis, among which posterior uveitis is the most common.[9,11] Other clinical manifestations are dysmyelination of nervous system, meningovascular diseases, cerebral abscesses and cerebral venous thrombosis.[3,7] Our patient was diag-

nosed with chronic meningoencephalitis associated with asymptomatic disc swelling accompanied with splinter hemorrhage and vasodilation of retinal veins in one eye and mild optic atrophy in the other eye simultaneously, a rare feature in neurobrucellosis. Since CSF opening pressure was normal, ophthalmoscopic findings were best diagnosed as papillophlebitis. Nevertheless, neglected raised intracranial pressure (RICP) leading to undiagnosed bilateral papilledema should be kept in mind, similar to a case reported by Nooshin B *et al.*[10] Papillophlebitis is a rare condition typically seen in young adults characterized by unilateral optic disc swelling in association with marked retinal venous engorgement resulting in merely mild diminished visual acuity. Appearance of the fundus in papillophlebitis is similar to that of central retinal vein occlusion (CRVO), so the term papillophlebitis has been used for healthy young patients instead of CRVO.[8] Pathogenesis of ophthalmic brucellosis has been proposed to be direct presence of brucella and immune complexes and meningeal inflammation leading to flow change and axonal degeneration.[9] Outcome of papillophlebitis is generally good without visual sequelae and the fundus appearance will be returning to normal.[13] Follow up fundus examination are recommended to ensure its stabilization and subsequent remission. Treatment of non-infectious papillophlebitis with steroids or blood thinning had been attempted. Although the disease is usually self-limiting, but systemic steroid has a beneficial effect. Our patient, however, was treated with antibiotics plus dexamethasone after which clinical improvement had been achieved.

CONCLUSION

We conclude that neurobrucellosis, this curable infectious disease though with grave morbidity and even mortality if overlooked, should be considered in any patient living in endemic areas, whether or not presenting with unusual symptoms.

CONFLICT OF INTEREST

None.

REFERENCES

1. Pappas G, Akritidis N, Bosilkovski M, Tsianos E. Brucellosis. N Engl J Med 2005; 352: 2325-36
2. Zeinalian Dastjerdi M, Fadaei Nobari R, Ramazanpour. Epidemiological features of human brucellosis in central Iran, 2006-2011. J Public Health. 2012 Dec;126(12):1058-62.
3. Hussein Algahtani, Bader Shirah et al. Neurobrucellosis Mimicking a Brain Tumor: A Case Report and Review of the Literature. Case Reports in Infectious Diseases, Volume 2017, Article ID 1434051, 5 pages.
4. A. S. Fauci, E. Braunwald, D. L. Kasper et al., Eds., Harrison's Principles of Internal Medicine, chapter 194e, McGraw-Hill Education, New York, NY, USA, 19th edition, 2015.
5. Stranjalis G, Singounas E, Boutsikakis I, Saraglou G. Chronic intracerebral Brucella abscess. J Neurosurg 2000; 92: 189.
6. Tumer Guven, Kenan Ugurlu et al. Neurobrucellosis: Clinical and Diagnostic Features.
Clinical Infectious Diseases, Volume 56, Issue 10, 15 May 2013, Pages 1407–1412,
7. Santini C, Baiocchi P, Berardelli A, Venditti M, Serra P. A case of brain abscess due to Brucella melitensis. Clin Infect Dis 1994; 19: 977-8.
8. Purvin VA. Optic neuropathies for the neurologist. Semin Neurol 2000; 20: 97–110.
9. Rolando I, Olarte L, Vilchez G, Lluncor M, Otero L, Paris M, Carrillo C, Gotuzzo I. Ocular Manifestations Associated with Brucellosis: A 26-year Experience in Peru. Clin Infect Dis. 2008; 46: 1338–45.
10. Nooshin Bazzazi, Alireza Yavarikia et al. Ocular Involvement of Brucellosis. Middle East Afr J Ophthalmol. 2013 Jan-Mar; 20(1): 95–97.
11. Rabinowitz R, Schneck M, Levy J, Lifshitz T. Bilateral Multifocal Choroiditis with Serous
Retinal Detachment in a Patient with Brucella Infection: Case Report and Review of the literature. Arch Ophthalmol. 2005; 123: 116–8.
12. Rolando I, Vilchez G et al. Brucellar uveitis: intraocular fluids and biopsy studies. Int J Infect Dis. 2009; 13: 206–11.
13. DukerJS, Sergott RC, Savino, PJ, Bosley TM. Optic neuritis with secondary retinal venous stasis. Ophthalmology. 1989; 96: 475-48.