Neurobrucellosis Manifesting as an Altitudinal Visual Field Defect

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ABSTRACT

Brucellosis is a zoonotic disease with a wide range of ophthalmic manifestations, including optic neuritis. It is most commonly caused by the consumption of raw or unpasteurised dairy products. We report a case of a 43-year male patient with an altitudinal visual field defect and unilateral optic neuritis caused by *Brucella*. While his visual acuity improved within 6 months of receiving anti-*Brucella* therapy and corticosteroid therapy, the altitudinal visual field defect persisted. Brucellosis can affect the central nervous system and eyes, in addition to having systemic manifestations. The ocular involvement of *Brucella* remains poorly understood. Early diagnosis and appropriate treatment can prevent serious complications in endemic areas.

Key Words: *Optic neuritis, Neurobrucellosis, Ocular brucellosis.*

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INTRODUCTION

Brucellosis is a zoonotic disease that is caused by *Brucella* species with multiple biotypes. Annually, 500,000 new cases of brucellosis are reported worldwide.[1] *Brucella* is transmitted to humans through the consumption of unpasteurised dairy products or uncooked meat or through contact with the skin, blood, or conjunctiva. It enters the reticuloendothelial system and settles in the lymph nodes, spleen, liver and bone marrow.

Central nervous system (CNS) involvement has been reported in 5–7% of patients with brucellosis. Optic nerve and chiasmatic involvement is extremely rare in patients with neurobrucellosis.[2] Optic nerve involvement is considered to be secondary to ocular or meningeal inflammation, and changes in the optic nerve follow axonal degeneration secondary to inflammation caused by brucellosis.[3] The expected ocular presentations in patients with brucellosis are conjunctivitis, episcleritis, scleritis, dacryoadenitis, granulomatous or non-granulomatous uveitis, optic neuritis, and papilloedema. These symptoms have also been reported in patients with ocular brucellosis.[4]

Here, we present a case of a patient with brucellosis and describe the aetiology of altitudinal visual field defects.

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CASE REPORT

A 43-year male patient was admitted to our emergency room because of sudden painless vision loss. The patient was evaluated by an ophthalmologist. Optic neuritis was detected in his left eye, and he was referred to the neurology clinic for diagnostic assessment. The patient was hospitalised for the differential diagnosis of optic neuritis.

The patient, who had chronic kidney failure and had undergone peripheral bypass surgery, did not use any drug other than 100 mg cilostazol 2 × 1 per oral (p.o.). He did not have a fever. His neurological examination was normal, except for vision loss. The visual loss was not accompanied by pain, redness, or any limitation in eye movements. His decimal best-corrected visual acuity was 1.0 in the right eye and 0.2 in the left eye. A relative afferent pupillary defect was detected in the left eye. Fundus examination showed that his left papillary margins were swollen and hyperaemic. Perimetric examination revealed an inferior altitudinal visual field defect in the left eye. The left optic disc was stained by fundus fluorescein angiography (Figure 1).

The findings of cranial magnetic resonance imaging (MRI) were normal. Orbital MRI revealed increased enhancement of the left optic nerve. The findings of echocardiography and carotid–vertebral Doppler ultrasonography were normal. Detailed tests were then performed for diagnosis. The results of the Venereal Disease Research Laboratory (VDRL) test, *Treponema pallidum* haemagglutination (TPHA) assay, hepatitis B surface antigen (HBsAg) test, hepatitis C antibody (anti-HCV) test, human immunodeficiency virus antibody (anti-HIV) test and purified protein derivative (PPD) skin test were negative. Moreover, the patient was negative for *Brucella* IgM but positive for *Brucella* IgG. The result of the Coombs anti-*Brucella* test was positive with a 1:640 titre. The patient had a history of eating

cheese made from unpasteurised raw milk in the countryside. He was evaluated for systemic infectious diseases; there was no evidence of past or current systemic involvement of brucellosis. Lumbar puncture revealed a slight pleocytosis in the cerebrospinal fluid (CSF) with mature white blood cells, predominantly lymphocytes. The results of *Brucella* tube agglutination, slide agglutination and tube agglutination with Coombs antiserum tests were negative in the CSF samples. Moreover, the blood and CSF cultures were negative.

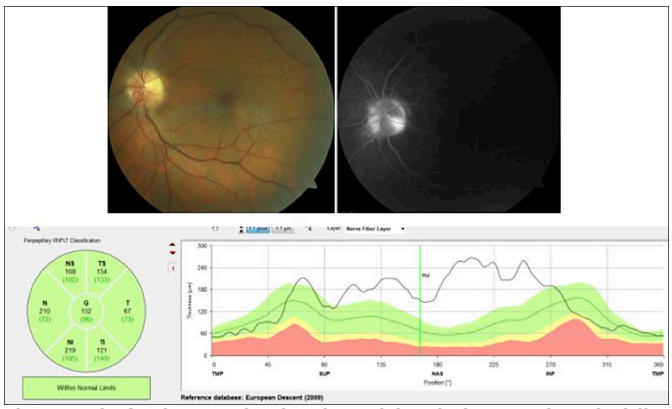

Figure 1: The fundus examination showed that the hyperemic optical disc margins of the papilla on the left were swollen and hyperemic. The retinal nerve fibre layers were observed thick in all the quadrants. Fundus fluorescein angiography was stained on the optic disc on the left.

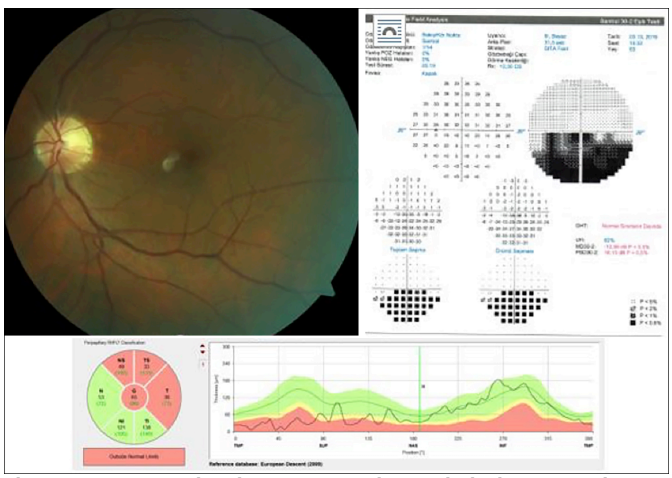

Figure 2: It was seen that there was an optic atrophy in the temporal, superior temporal and superior nasal quadrants on the left eye and altitudinal inferior visual field loss after the six-months.

The cause of vision loss was evaluated for brucellosis-related optic neuritis. Due to the presence of pleocytosis in the CSF, the diagnosis was accepted to be neurobrucellosis despite the absence of neurological findings.

Antibiotic therapy was initiated, including doxycycline (200 mg/day p.o.), rifampicin (600 mg/day p.o.) and ceftriaxone (2 g/day intravenous [i.v.]). The treatment duration was 6 weeks. Because of optic nerve involvement, methylprednisolone was started at 1 mg/kg/day and continued for 2 weeks. Following this, the dose of prednisone was tapered; prednisone was discontinued by decreasing 8 mg every 5 days in the following period. Corticosteroid therapy was discontinued after 2 months.

At the 6-month follow-up, the *Brucella* serum agglutination titre decreased to 1:20. The decimal best-corrected visual acuity was 1.0 in the right eye and 0.8 in the left eye. Optic atrophy and an altitudinal visual field defect were detected on the left side (Figure 2).

DISCUSSION

We present a case of neurobrucellosis with only ocular symptoms and no additional systemic findings. Cranial nerve involvement, including vestibulocochlear, trigeminal, facial, abductive and oculomotor nerves, has been reported in patients with neurobrucellosis. Optic nerve involvement is uncommon in these cranial neuropathies.[5]

A definitive diagnosis of neurobrucellosis was not possible in our patient as the cultures were negative; however, we made the diagnosis on the basis of a history of consuming unpasteurised milk or milk products in an endemic country, the exclusion of other clinically significant aetiologies in this setting, a *Brucella* serum agglutination titre >1:160, abnormal CSF findings suggestive of CNS inflammation and the improvement in visual acuity with appropriate treatment. Marques *et al*. diagnosed neurobrucellosis on the basis of a history of consuming unpasteurised dairy products, a *Brucella* serum agglutination titre of 1:640 and pleocytosis in the CSF in a patient who was admitted with acute visual impairment in the right eye.[6] It is difficult to isolate *Brucella* species in blood and CSF cultures. In a retrospective series involving five patients with neurobrucellosis, the serum culture was positive in one patient and the CSF culture was positive in another patient; however, *Brucella* species could not be isolated from the blood and CSF of the remaining patients.[7]

Ocular involvement types have been found to vary in patients with systemic brucellosis. Rolando *et al*. reported that 52 out of 1551 patients with brucellosis had ocular brucellosis.[3] Of these 52 patients, 43 (82.7%) had uveitis, with the most common uveal syndrome being posterior uveitis. Optical disc oedema was observed in five patients (9.6%). Of the 52 patients with ocular brucellosis, 15 (28.8%) had no systemic symptoms.

In a study conducted in Turkey, Sungur *et al.* reported that 28 (21%) out of 132 patients with brucellosis had ocular involvement.[8] In that study, all patients with anterior uveitis were in the acute phase, while those with choroiditis, papillary oedema, and retinal haemorrhage were in the chronic phase of the disease.

Ocular manifestations of brucellosis can be observed in the chronic phase of the disease. There was no systemic evidence of brucellosis, except for optic neuritis, in the present patient, who was evaluated in the chronic phase of the disease. The only clinical findings in this patient were left-sided optic neuritis and pleocytosis in the CSF with Brucella IgG positivity.

Neurobrucellosis has a wide range of clinical manifestations; however, optic nerve involvement manifesting as an altitudinal visual field defect is quite rare. As evident from the present case, at times, optic neuritis might be the only symptom of brucellosis.

Therefore, to prevent severe loss of vision, ocular involvement of brucellosis should be taken into consideration, especially in endemic regions, while investigating the aetiology of optic neuritis.

CONFLICT OF INTERESTS:

The authors declared no competing interest.

PATIENT'S CONSENT:

Informed consent was taken from the patient for publication of this case report and related images.

AUTHORS' CONTRIBUTION:

GGU: Data collection, literature review, and writing the manuscript.
SYB: Assistance in manuscript designing and data collection.
HT: Supervision, and critical revision.
All the authors have approved the final version of the manuscript to be published.

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