

Unilateral optic neuritis as a presentation of neurobrucellosis

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Abstract

Neurobrucellosis manifesting as optic neuritis is a rare disease in childhood. We report a case of neurobrucellosis in a 11 year old girl leading to visual impairment and headache. Physical examination revealed mild oedema of right tibiotarsic joint and optic neuritis. Investigations showed CSF pleocytosis and a Brucella serum agglutination titer of 1/640. Complete reversal of the symptoms was observed after appropriate antibiotic treatment. To our knowledge only four cases of neurobrucellosis manifesting with visual impairment in childhood are previously reported in literature.

Introduction

Brucellosis is a zoonosis disease, well-documented cause of fever of unknown origin, with

varied and nonspecific symptoms. Involvement of the central nervous system (CNS) is a rare occurrence in childhood and is present in less than 2% of cases, predominantly with *B. Melitensis*. The clinical presentation of brucellosis localized to CNS is diverse and localized forms are difficult to diagnose. There are a few reports concerning cranial nerves involvement.^{1,2}

The available literature on optic neuritis in childhood stems largely from case report or retrospective case series. Optic neuritis presents differently in children than in adults. It is considerable less common in children and most cases are due to an immune-mediated process following an antecedent viral illness.³ Optic neuritis has also been linked to acute infection with Epstein Barr Virus, *Borrelia*, measles, mumps and varicella. Less commonly, optic neuritis may be the first manifestation of multiple sclerosis. Optic neuritis linked to *Brucella* infection is rare and scarcely reported in literature. We report a case of childhood neurobrucellosis presenting as optic neuritis.

Case Report

A previously healthy 11 year old girl was admitted with headache, retroorbital pain and acute right eye visual impairment. She had no significant personal or family history, except for a vacation in Portuguese countryside and a consumption of unpasteurized dairy products. There wasn't any recent immunization or preceding infection. Physical examination revealed sub-febrile temperature (37.7°C), mild oedema of right tibiotarsic joint, afferent

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pupillary defect and a funduscopy with optic disc swelling. Best corrected visual acuity was 20/30 (Snellen chart) in the right eye. Other ocular lesions weren't reported. Neurologic examination was normal. Initial diagnostic was a post-infectious optic neuritis so endovenous methylprednisolone (250 mg, 6/6h) was initiated. Investigations showed a Brucella serum agglutination titer of 1/640. A lumbar puncture disclosed a slight pleocytosis of the cerebrospinal fluid, with mature white blood

Table 1. Literature review for neurobrucellosis optic neuritis.

Studies	Tali <i>et al.</i> ¹⁰	Tonekaboni <i>et al.</i> ⁵	Karapinar <i>et al.</i> ¹¹	Elrazak ¹²	Our case
Age gender	8 year-old boy	13 years-old boy	15 years-old girl	13 years-old girl	11 years-old girl
Brucella background and treatment received	Yes Streptomycin-tetracycline combination for 15 days	Yes Rifampin + doxycycline for 2 months + streptomycin for 25 days	No	No	No
Clinical manifestation	Diplopia, gradual loss of vision, facial paraesthesiae, vertigo, headache	Visual impairment and unilateral hearing loss	Acute bilateral blindness	Acute blindness of the left eye	Headache, retroorbital pain and acute right eye visual impairment
Antibiotic treatment	Rifampicin for 6 months	Doxycycline + rifampin + trimethoprim-sulfamethoxazole + ciprofloxacin for 1 year	Rifampicin + doxycycline for 3 months + streptomycin for 1 month	Tetracycline for 2 months + streptomycin for 3 weeks	Doxycycline + Rifampicin for 6 months
Corticoid treatment	No	Prednisolone for 2 months	Methyl prednisolone for 7 days	Prednisolone for 3 months	Methyl prednisolone for 1 day + Prednisolone for 1 month
Resolution of symptoms	The headache, diplopia, vertigo and paraesthesiae had resolved, but the scotoma persisted	Visual acuity, but not hearing, improved	Complete recover	Complete recover	Complete recover

cells, predominantly lymphocytes. PCR for *Brucella* in the CSF was negative and MRI scan was normal. Oligoclonal bands were not performed. Serum samples tested negative for mycoplasma, cytomegalovirus, Epstein Barr and toxoplasmosis. Rheumatological profile (rheumatoid factor, ANA, ANCA, C3, C4) was normal.

Antibiotic therapy with Doxycycline (200 mg/d) and Rifampicin (600 mg/d) was performed during 6 months. Complete reversal of the visual and systemic symptoms was observed, with a 12 month follow-up. A decrease in *Brucella* serum agglutination titer to 1/80 was observed. Audiologic studies were performed and were normal.

Discussion

A definitive diagnosis of Neurobrucellosis is not possible as cultures were negative, but we based our diagnostic in a history of ingesting untreated milk or milk products in an endemic country, exclusion of other clinically significant etiologies in this setting, a serum agglutination titer for *Brucella* greater than 1:160, abnormal cerebrospinal fluid findings suggesting CNS inflammation, and resolution of symptoms with appropriate treatment.

Neurobrucellosis has neither a typical clinical picture nor specific cerebrospinal fluid findings.^{4,5} *Brucella* organisms are fastidious

and difficult to isolate in blood and CFS cultures, so serological tests are important.^{6,7}

There are few reports in literature regarding optic neuritis as a manifestation of neurobrucellosis in children. In the literature reviewed we found 4 case reports that are presented in Table 1. Treatment of acute brucellosis in adults and children older than 8 years, World Health Organization (WHO) guidelines recommend rifampin (600-900 mg) and doxycycline (200 mg) daily for a minimum of 6 weeks. Duration of therapy is controverse. We have chosen a 6 month therapy, as we were concerned on the possibility of relapse, especially if treatment is short.⁸

In conclusion, in the investigation of acute optic neuritis in endemic countries, as the Mediterranean and the Middle East ones, it is important the exclusion of this disease, as it is a treatable cause.⁹

References

1. Akdeniz H, Irmak H, Anlar O, Demiroz AP. Central nervous system brucellosis: presentation, diagnosis and treatment. *J Infect* 1998;36:297-301.
2. Al-Sous MW, Bohlega S, Al-Kawi MZ, et al. Neurobrucellosis: Clinical and Neuroimaging Correlation. *AJNR* 2004;25: 395-401.
3. Morales DS, Siatkowski RM, Howard CW, Warman R. Optic neuritis in children. *J Pediatr Ophthalmol Strabismus* 2000;37: 254-9.
4. McLean DR, Russell N, Khan MY. Neurobrucellosis: clinical and therapeutic features. *Clin Infect Dis* 1992;15:582-90.
5. Tonekaboni S., Karimi A., Armin S., Khase L.A., Sabertehrani A.S. Neurobrucellosis: A Partially Treatable Cause of Vision Loss. *Pediatric Neurology*, 2008; 40: 401-403.
6. Omar F, Zuberi S, Minns R. Neurobrucellosis in childhood: six new cases and a review of the literature. *DMCN* 1997;39: 762-5.
7. Solomon HM, Jackson D. Rapid diagnosis of *Brucella melitensis* in blood: some operational characterizations of the BACT/ALERT. *J Clin Microbiol* 1992;30:222-4.
8. Bouza E, Torre MG, Guerrero A, et al. *Brucella* meningitis. *Rev Infect Dis* 1987;9: 810-82.
9. Pappas G, Akritidis N, Bosilkovski M, Tsianos E. Brucellosis. *N Engl J Med*, 2005; 352:2325-36.
10. Tali ET, Keskin T, Oznur II, Simonson T, Yuh WT. MRI of *Brucella* polyneuritis in a child. *Neuroradiology*, 1996; 38(Suppl.1): 190-2.
11. Karapinar B, Yilmaz D, Vardar F, et al. Unusual presentation of brucellosis in a child: Acute blindness. *Acta Paediatrica*, 2005;94:378-80.
12. Elrazak A. *Brucella* optic neuritis. *Arch Intern Med* 1991;151:776-8.