Case Report

Inflammatory choroidal neovascular membrane in presumed ocular Lyme borreliosis

Radgonde Amer,¹ Suzanne Brannan² and John V. Forrester¹

¹Department of Ophthalmology, Aberdeen Royal Infirmary, UK
²Department of Ophthalmology, Queen Margaret Hospital, Dunfermline, UK

ABSTRACT.
Introduction: Lyme disease is a multisystemic disease with protean ocular manifestations. We describe the occurrence of inflammatory choroidal neovascular membrane (CNVM) in two patients suffering from presumed Lyme disease.

Methods: Descriptive review of the clinical records of two patients.

Results: Patient 1: 16-year-old healthy male presenting with a visual acuity of counting fingers [oculus dexter (OD)] and 6/6 [oculus sinister (OS)] 3 months after a tick bite. He had papillitis and an exudative subretinal macular lesion OD. Treatment was started with intravenous (IV) ceftriaxone; a week later, IV methylprednisolone was administered with a tapering dose of oral steroids thereafter. Three months later, VA had improved to 3/60 OD. Patient 2: 38-year-old healthy female presenting with reduced left-eye vision (6/24) 6 weeks after a tick bite. She also suffered from erythema migrans and arthralgias. She had left-eye papillitis, macular haemorrhages and vascular sheathing. Treatment was started with IV ceftriaxone. One month later, there was profound loss of vision with development of CNVM. Treatment was declined by the patient and eventually retinal fibrosis developed.

Conclusion: Inflammatory CNVM has not been described previously in the setting of ocular Lyme borreliosis. We herein describe the occurrence of inflammatory CNVM in two patients whose diagnosis with Lyme disease was clinically based – both were sero-negative. Visual outcome in the two patients was profoundly impaired because of the ensuing macular scar.

Key words: choroidal neovascular membrane – Lyme disease – maculopathy – uveitis

© 2008 The Authors
Journal compilation © 2008 Acta Ophthalmologica

doi: 10.1111/j.1755-3768.2007.01160.x

Introduction

Lyme disease is a multisystemic disease, caused by the tick-borne spirochaete Borrelia burgdorferi. The spectrum of ocular borreliosis includes episcleritis, interstitial keratitis, anterior uveitis, vitritis, choroiditis, retinal vasculitis, optic neuritis and papilloedema (Mikkila et al. 2000). We herein report on the occurrence of inflammatory choroidal neovascular membrane (CNVM) in two patients suffering from presumed Lyme disease.
haemorrhages lining the walls of the blood vessels in the nasal and temporal peripheral retinas bilaterally, more marked temporally. Fluorescein angiogram revealed papillitis and active CNVM surrounded by subretinal fluid OD. There was evidence of active vasculitis both in the posterior pole and in the periphery of the right eye (Fig. 1B). Similar vasculitis was observed in the peripheral retinal vessels of the left eye. Lyme serology [enzyme-linked immunosorbent assay (ELISA) and western immunoblot assay] was negative, as was syphilis serology. Treatment was started with a 2-week course of intravenous (IV) ceftriaxone (2 g/day); 1 week later, he received a 3-day course of IV methylprednisolone (1 g/day), followed by a tapering regimen of oral steroids. Three months later, VA had improved to 3/60 OD with a residual macular fibrotic lesion and no signs of active vasculitis in either eye.

Case 2
A 38-year-old healthy female presented to our clinic with reduced vision in her left eye 6 weeks after being bitten by a tick. She mentioned that this happened in mid-June on the west coast of Scotland and that the tick was removed the following day. The bite site developed swelling and erythema; within a week, she suffered from a fleeting macular rash of both legs. She had arthralgia, mainly of her knees and fingers. On presentation, VA was 6/6 OD and 6/24 OS. She was on doxycycline tablets (100 mg/day) that were prescribed to her 1 week before her presentation to us. She had moderate anterior chamber reaction, no keratic precipitates and few snowballs in the inferior vitreous. On fundoscopy she had a swollen optic disc, small retinal haemorrhages in the centre of the macula and a dot-like yellow foveal lesion. A sheathed retinal vessel was seen just inferior to the haemorrhages. No perivascular sheathing was noted elsewhere. Her right eye did not show any signs of inflammation. Work-up for possible infectious aetiology, including syphilis, was uneventful. Lyme serology (ELISA) was negative. Based on the highly characteristic presentation, it was decided to start treating her for presumed Lyme disease. Treatment commenced with a 4-week course of intravenous ceftriaxone at 2 g/day, following which she noted a reduction in her central scotoma on Amsler grid. Unfortunately, vision remained unchanged. A month later, she experienced further reduction in vision with VA dropping to counting fingers. There was enlargement of the yellow foveal lesion with the appearance of a surrounding subretinal fluid and retinal striae. Further treatment was declined by the patient. Marked retinal fibrosis eventually developed in the centre of the foveal lesion (Fig. 2). Serological test remained negative.

Discussion
Inflammatory CNVM has not been described previously in the setting of ocular Lyme borreliosis. We describe the occurrence of inflammatory CNVM in two patients whose diagnosis with Lyme disease was clinically based – both were sero-negative. Both patients suffered from a tick bite in an endemic area of Scotland and both patients reported extracting the tick (in one patient it was delayed until the following day). One patient had solely ocular involvement while the second
Fig. 2. Clinical photograph of the left fundus showing a subretinal yellow macular lesion with a marked overlying retinal fibrosis and retinal striae extending nasally. Small deep retinal haemorrhages are noted on its infero-temporal edge.

patient developed the characteristic erythema migrans rash with arthralgia. Sero-negative Lyme is an established entity that may be attributed to sequestration of the spirochetes into immunologically privileged sites such as the central nervous system or the interior of the eye (Steere 1989). Previous reports described impaired immune response in uveitis patients with confirmed diagnosis of Lyme disease (Dattwyler et al. 1988).

The ophthalmic manifestations of Lyme borreliosis represent a spectrum that includes ocular inflammatory conditions (conjunctivitis, episcleritis, interstitial keratitis, uveitis and retinal vasculitis), neuro-ophthalmic involvement (cranial neuropathies such as facial nerve palsy, optic neuritis and optic disc oedema) and orbital pathology (orbital myositis) (Mikkila et al. 2000). Many of these clinical features are also characteristic of syphilitic eye involvement. A histological study of four eyes with syphilitic chorioretinitis (Blodi & Hervouet 1968) showed characteristic dehiscences in Bruch’s membrane leading to the invasion of the choroid by retinal elements namely retinal glia or pigment epithelium. Haematogenous spirochetal dissemination and the accompanying inflammatory process could have led to breaks in Bruch’s membrane through which CNVM developed. The marked retinal fibrosis overlying the CNVM seen in case 2 suggests that proliferation of the retinal glial tissue is actively attempting to contain – and thus limit – the spirochetal invasion into the retina.

*Borrelia burgdorferi* sensu lato is the spirochete responsible for Lyme disease, but isolates are phenotypically and genotypically heterogeneous (Wang et al. 1998; Logan 1994; Postic et al. 1998). The different genomic groups are associated with different geographical areas. Ling et al. (2000) characterized 12 *B. burgdorferi* sensu lato isolates cultured from ticks collected in the Highlands of Scotland. The presence of different strains in Scotland compared with other areas of Europe, and different strains within one small area of Scotland, is comparable to many previous studies that have also identified strain heterogeneity within these genomic groups (Wang et al. 1998; Mathiesen et al. 1997). This fact may be directly responsible for the protean ophthalmic manifestations of Lyme disease and suggests that the development of inflammatory CNVM might be a sequela to a specific genomic type of *B. burgdorferi* in Scotland.

Visual outcome in the two patients was impaired profoundly because of the ensuing macular scar, while the concurrent retinal vasculitis and papillitis showed total resolution with the antibiotic therapy. Direct ocular infection and a delayed hypersensitivity mechanism may be involved at different disease stages (Bodaghi 2007). This highlights the need to evaluate combination therapies in the treatment of ocular Lyme borreliosis with antibiotics and anti-inflammatory agents to control the resultant inflammatory response.

**References**


Received on August 17th, 2007.
Accepted on November 8th, 2007.

**Correspondence:**
Radgonde Amer
Department of Ophthalmology
University of Aberdeen
Foresterhill
AB25 2ZD
UK
Tel: + 44 1224 533 782
Fax: + 44 1224 555 955
Email: radgondeamer@yahoo.com