Q Fever Endocarditis with Bilateral Multifocal Retinitis: A Case Report

Rym Maamouri¹, Olfa Beizig¹, Khadija Mzoughi², Monia Cheour²

¹Department of Ophthalmology, Hospital Habib Thameur, Tunis, Tunisia, ²Department of Cardiology, Hospital Habib Thameur, Tunis, Tunisia

Abstract

Purpose: To describe a case of bilateral retinitis in a patient with endocarditis and a serologically confirmed Q fever.

Methods: A single case report documented with multimodal imaging.

Results: A 55-year-old patient with culture-negative endocarditis was referred to our department for an ocular examination. His visual acuity was 20/20 in both eyes. Fundus examination showed white retinal infiltrates with few superficial retinal hemorrhages scattered in the posterior pole. There was no staining on fluorescein angiography. Swept-source optical coherence tomography (SS-OCT) revealed increased inner retinal reflectivity with a focal area of retinal thickening. Laboratory tests showed a high titer of antibodies against *Coxiella burnetii*. The patient was treated with doxycycline. Two weeks later, fundus examination showed partial resolution of retinitis with inner retinal thinning in SS-OCT.

Conclusions: Multifocal retinitis is an uncommon presentation of Q fever. The diagnosis should be considered, especially when associated with culture-negative infective endocarditis, highlighting the importance of routine ocular examination.

Keywords: Coxiella burnetii, Endocarditis, Q fever, Retinitis

Address for correspondence: Rym Maamouri, 3, Rue Ali Ben Ayed, Montfleury, Tunis 1089, Tunisia.

E-mail: ryma.maamouri@fmt.utm.tn

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INTRODUCTION

Q fever, first described in 1937, is a ubiquitous zoonosis caused by an intracellular bacterium, *Coxiella burnetii* (CB) with a worldwide distribution characterized by clinical polymorphism.¹ Systemic infection can take two forms: the acute disease, which is more frequent and often asymptomatic and the chronic form with a blood-culture negative endocarditis as the most frequent and serious presentation.^{2,3} Herein, we report a case of bilateral multifocal retinitis associated with serologically confirmed Q fever.

CASE REPORT

A previously healthy, 55-year-old farmer was referred to the cardiology department for the evaluation of a persistent fever for 3 months and aortic valve murmur. He was otherwise

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asymptomatic and denied any drug use. A general examination showed no cutaneous manifestations. He presented a biological inflammatory syndrome. Microbial investigation with several and repeated blood cultures remained negative. Transthoracic and transesophageal echocardiography showed mild aortic regurgitation and vegetation on the aortic valve that measured 7 mm × 16 mm [Figure 1].

The diagnosis of infective endocarditis on the native aortic valve was possible based on *modified Duke* criteria (one major and one minor).⁴

Therefore, the patient was referred to the ophthalmology department for an opinion. On examination, his best-corrected visual acuity was 20/20 in each eye (OU). He had no relative afferent pupillary defect and a full extraocular

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motility OU. On slit-lamp examination, no inflammatory cells were observed in the anterior chamber or the vitreous in OU. Fundus examination revealed small white retinal infiltrates with few superficial retinal hemorrhages scattered in the posterior pole OU [Figure 2a and b]. Fluorescein angiography (FA) [Figure 2c and d] showed no staining of the retinal infiltrates and a masking effect by retinal hemorrhages. Swept-source optical coherence tomography (SS-OCT) scans throughout the lesions revealed increased foci of thickening inner retinal reflectivity with sparing of outer retinal layers, retinal pigment epithelium, and choroid [Figure 3a and b].

Due to the presence of acute multifocal retinitis associated with possible infective endocarditis with negative cultures, we decided to complete diagnostic tests including *Brucella*, *Bartonella*, CB, rickettsia serology, and syphilis. The diagnosis of CB was confirmed by the detection of high titers of anti-CB immunoglobulin G (IgG) with the indirect immunofluorescent assays (IgG phase I: 128/phase II: 64 and IgM negative). Otherwise, all other serology tests returned negative. The patient was treated with oral doxycycline 100 mg twice a day with clinical and biological improvement. Vegetation size decreased on echocardiogram controls.

Two weeks later, a fundus examination showed complete resolution of the white retinal lesion in the right eye, and partial resolution of the foci of retinitis with a new superficial hemorrhage in the left eye without visible

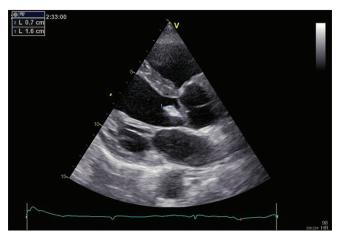


Figure 1: Transthoracic echocardiogram showing an aortic valve vegetation measuring 7 mm imes 16 mm

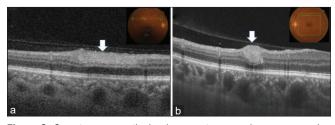


Figure 3: Swept-source optical coherence tomography scans passing through the retinal infiltrates showing increased internal reflectivity with retinal thickening (white arrow) respectively in the right (a) and left (b) eye

scars [Figure 4a and b]. SS-OCT scan showed a focal area of inner retinal atrophy corresponding to a resolved retinal infiltrate in the right eye [Figure 4c and d]. The patient was discharged after the improvement of his clinical condition. The consent for publication use of images was obtained from the patient.

DISCUSSION

Transmission of Q fever to humans occurs through inhalation

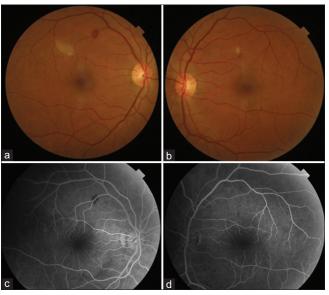


Figure 2: Fundus photographs showing superficial infiltrates at the posterior pole with perivascular hemorrhage respectively in the right (a) and left (b) eye. (c and d) Fluorescein angiography showed no staining of the retinal lesions with a masking effect by retinal hemorrhages

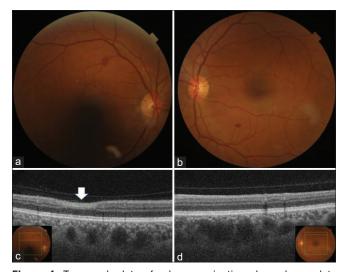


Figure 4: Two weeks later, fundus examination showed complete resolution of white retinal lesion in the right eye, partial resolution of the foci of retinitis with new superficial hemorrhage in the left eye without visible scars (a and b). Swept-source optical coherence tomography scan showed a focal area of inner retinal atrophy corresponding to a resolved retinal infiltrate in the right eye (c and d)

of infected aerosols, especially during calving in cattle, rarely through tick bites or ingestion of unpasteurized milk.² Infection in humans is often asymptomatic and frequently misdiagnosed by physicians. In the chronic phase, the general manifestations are mainly endocarditis, chronic fatigue syndrome, and rarely hepatitis.³ Endocarditis cases develop late, usually 2 months to 1 year following acute Q fever.⁵ Our findings are consistent with previous reports; our patient has developed endocarditis 3 months after a persistent fever.

Ocular involvement in Q fever is uncommon, but the most frequently reported impairment is neuro-ophthalmological manifestations, including optic neuritis and abducens nerve palsy.⁶ A case of exudative retinal detachment secondary to Q fever infection that resolved under doxycycline has been recently described.⁷ However, retinitis is extremely infrequent.^{8,9}

The first case was reported in a 41-year-old male returning from Mexico with fever and flu symptoms without endocarditis, unilateral multifocal retinitis, and positive serology. The second case was about a 67-year-old cattle breeder presenting with persistent fever, floaters, and endocarditis with a positive serology. Ocular findings were consistent with multifocal retinitis, retinal hemorrhage, and like hyper-reflective area in the middle retina at the OCT consistent with unilateral paramacular acute middle maculopathy. The retinal infiltrates were slightly hypofluorescent in FA. In both cases, the patients were treated with doxycycline and ocular improvement. Page 18.

To the best of our knowledge, our report is the third case of retinitis associated with a serologically proven CB infection described in the literature. SS-OCT remains an important tool and shows characteristic features in Q fever with the presence of a focal area of retinal hyperreflective thickening in the retinal nerve fiber layer corresponding to the retinal infiltrates which ultimately disappears clinically but leaves an inner retinal thinning at the OCT,⁹ as it was the case in our patient in the clinical course and at the SS-OCT. CB has a vascular tropism, the foci of superficial retinitis may result from intraretinal multiplication of the bacteria, or an immune-mediated response to bacterial antigens following deposition of immune complexes, inflammatory cells, or antibodies in retinal vessels.¹⁰

Serology remains the main diagnostic tool for Q fever. Indirect immunofluorescence tests are considered the gold standard for the diagnosis. ¹¹ In our case, the serological diagnosis was made by a high titer of IgG, IgM antibodies were negative because of delays in clinical suspicion thus confirming the chronic phase of the disease.

Prompt referral of patients with endocarditis to ophthalmologists is highly recommended. Making the correct

and oriented diagnosis of Q fever in these patients mainly with culture-negative endocarditis presenting with multifocal retinitis and superficial hemorrhage by the ophthalmologist remains crucial. Keep in mind that the prevention of this disease is possible by testing the cattle for serology and vaccinating those who are seronegative by involving the veterinarians preferably before the calving period.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that his name and initials will not be published and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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