BARTONELLA QUINTANA–ASSOCIATED NEURORETINITIS: LONGITUDINAL SPECTRAL-DOMAIN OPTICAL COHERENCE TOMOGRAPHIC FINDINGS

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Purpose: To report an unusual case of neuroretinitis caused by *Bartonella quintana* and its spectral-domain optical coherence tomographic (SD-OCT) features.

Methods: A 12-year-old girl presented with unilateral neuroretinitis with stellate maculopathy. Bartonellosis was confirmed after serologic testing for antibodies to *B. quintana*.

Results: Color photograph of the right eye revealed papillitis and stellate macular exudation. spectral-domain optical coherence tomography of the right eye revealed hyperreflective dots in the outer nuclear and outer plexiform layers, as well as disruption and loss of the external limiting membrane, ellipsoid zone, and interdigitation zone in the foveal area.

Conclusion: The authors report an unusual case of neuroretinitis by *B. quintana* and its spectral-domain optical coherence tomographic findings.

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Bartonella is a gram-negative facultative intracellular bacteria with more than 30 species described as a human or an animal pathogen.[1–3] Bartonellosis or cat scratch disease is transmitted to humans through a cat scratch or bite. Ticks and fleas have also been described as a vector for human infection by *Bartonella*. The main species related to human bartonellosis are *Bartonella henselae*, *Bartonella quintana*, and *Bartonella bacilliformis*.[1] Bartonellosis may present in a wide spectrum of systemic manifestations, including endocarditis, hepatitis, encephalopathy, meningitis, and hemolytic anemia. Ocular complications are atypical, and neuroretinitis represents the majority of cases with fundus involvement.[4–8]

B. henselae is the most common *Bartonella* species infecting humans and is associated with cat scratch disease.[9] *B. bacilliformis* is transmitted by sandflies and is related to Carrión disease and verruga peruana.[4,9] *B. quintana* is one of the few *Bartonella* species where humans are considered to be the main reservoir and was commonly associated with trench fever during World War I and II.[2,6,10] Currently, *B. quintana* is a reemerging pathogen causing urban trench fever, which is associated with alcoholism and an immunocompromised state.[9]

Neuroretinitis is characterized by optic disc swelling with a complete or partial macular star formation and occurs in approximately 2% of patients with bartonellosis.[11,12] Approximately, two thirds of patients with neuroretinitis have positive serologic testing for *B. henselae*[13] and the disease often manifests as unilateral, painless, and sudden visual loss. *B. quintana*, *Bartonella grahamii*, and *Bartonella elizabethae* have also been found to be the causative agents of neuroretinitis.[14–16] The diagnosis of *Bartonella* neuroretinitis is made by clinical presentation and is confirmed with positive serologic analysis for *Bartonella.*

There have been several reports of *henselae* and non-*henselae* species of *Bartonella* causing neuroretinitis. However, we are aware of only three previous reports of neuroretinitis caused by *B. quintana.*[17–19] The purpose

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of this study was to describe the clinical evolution of a neuroretinitis caused by *B. quintana* using spectral-domain optical coherence tomography (SD-OCT).

Case Report

A 12-year-old girl presented to clinic complaining of low-grade fever, frontal headache, and decreased visual acuity in the right eye persisting for 1 month. She reported no other recent flu-like symptoms, such as malaise or chills, but confirmed recent contact with animals, including cats. The patient had an unremarkable previous medical and ocular history.

On ocular examination, the best-corrected visual acuity was counting fingers in the right eye and 20/20 in the left eye. Pupillary reactions, slit-lamp biomicroscopy of the anterior segment, and intraocular pressure were normal. Color photograph of the right eye revealed optic disc edema, macular exudation in a star configuration, and retinitis manifested as multifocal well-circumscribed peripapillary white lesions. Left fundus examination was unremarkable (Figure 1). The SD-OCT (Cirrus HD OCT; Carl Zeiss Meditec, Inc, Dublin, CA) of the right eye revealed hyperreflective dots (predominantly temporal to the fovea) in the outer nuclear and outer plexiform layers corresponding to the intraretinal exudates seen within the macula on color photographs. The SD-OCT also showed disruption and loss of the external limiting membrane, ellipsoid zone, and interdigitation zone in the foveal area (Figure 2).

Laboratory tests revealed elevation of C-reactive protein, anormocytic–normochromic anemia, normal leukocyte count with a slight right shift, and normal chest and sinus x-rays. Serologies for cytomegalovirus, herpes simplex virus, human immunodeficiency virus, *Histoplasma capsulatum*, *Toxoplasma gondii*, *Toxocara canis*, and *Borrelia burgdorferi* were negative. *B. quintana* indirect fluorescent antibody titers were positive for IgG at 1:512 but were negative for IgM. A *B. henselae* IgG antibody reaction (1:64) associated with a nonreactive IgM antibody was also observed.

Oral doxycycline at 100 mg twice daily and prednisone (0.5 $mg \cdot kg^{-1} \cdot day^{-1}$) were initiated. Two months after the treatment initiation, there was significant decrease in the amount of optic disc edema and macular exudation, and the visual acuity of the right eye improved to 20/70. However, the disruption and loss of the outer retinal layers persisted after treatment (Figure 2).

Discussion

An increasing number of *Bartonella* species have been associated with various reservoirs, vectors, and clinical manifestations. Cat scratch disease is one of the most well-known clinical presentations of *Bartonella* species infections, most of them caused by *B. henselae*.[1–3] Bartonellosis by *B. quintana* is a disease predominantly found in undomiciled, and several recent cases have been described in the literature. Parinaud oculoglandular syndrome and neuroretinitis are the most frequent ocular manifestations of bartonellosis and typically presents as a unilateral and painless decrease of vision in conjunction with optic disc edema and either a partial or complete macular star.[1–3]

In this case report, the diagnosis of bartonellosis was confirmed by patient history and clinical manifestations, positive *B. quintana* serology, and successful therapeutic response. The patient had contact with cats and complained of typical *Bartonella* infection symptoms before experiencing visual symptoms. A diagnosis of *B. quintana* was made after elevated IgG titers of 1:512 were found on indirect fluorescent antibody testing. Titers with values of >1:256 are considered strongly suggestive of recent infection.[20–22] Cross-reactivity with *B. henselae* IgG antibody probably occurred given that a low titer of this immunoglobulin (1:64) was identified in association with a nonreactive IgM antibody. In the period of two months of treatment with doxycycline, the patient's visual acuity improved, and there was a decrease in the neuroretinitis activity. The present case showed a classic *Bartonella* neuroretinitis characterized by papillitis and lipoidal star in the macula.

To our knowledge, there are only three other cases of neuroretinitis associated with *B. quintana.* These cases demonstrated similar clinical features of optic

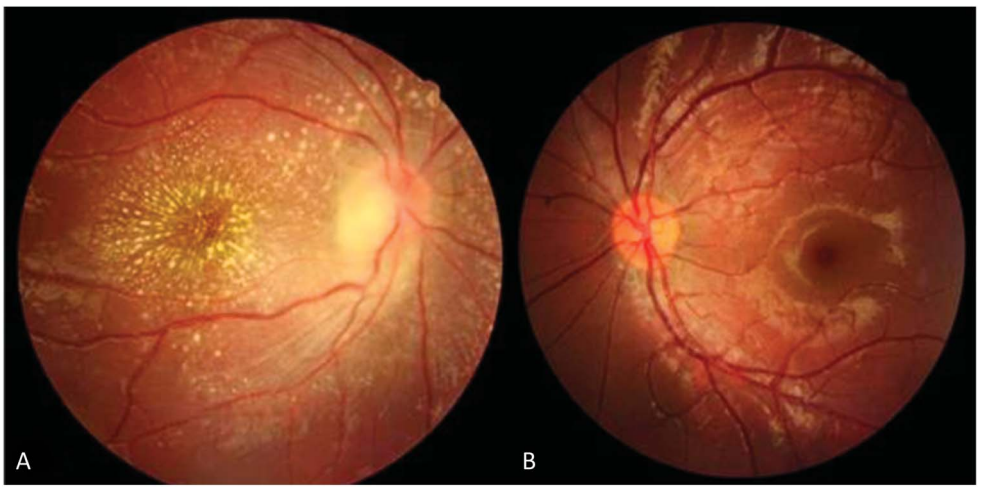

Fig. 1. A. Color photograph of the right eye showing papillitis, stellate macular exudation, and multifocal peripapillary white lesions. **B.** Color photograph of left eye was normal.

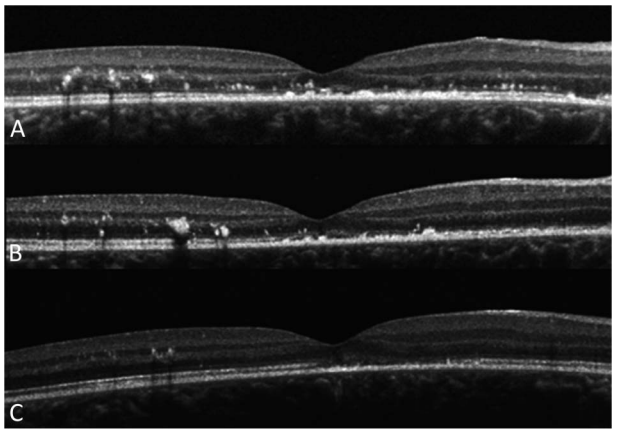

Fig. 2. A. Baseline SD-OCT of the right eye showed intraretinal exudates manifested as hyperreflective dots and disruption and loss of the external limiting membrane, ellipsoid zone, and interdigitation zone in the foveal area. **B.** At 60 days of follow-up, there was a decrease in the amount of intraretinal exudates with maintenance of the outer retinal layers disruption. **C.** At 6 months of follow-up, there was a minimal amount of intraretinal exudates and persistence of outer retinal layers disruption in the foveal area.

disc edema and macular exudation in a star configuration.[17–19] However, they did not mention the OCT changes caused by *B. quintana*. In the present report, SD-OCT showed hyperreflective dots in the outer nuclear and outer plexiform layers corresponding to the intraretinal exudates typically seen on neuroretinitis cases caused by *Bartonella*. In addition, the SD-OCT revealed disruption and loss of the external limiting membrane, ellipsoid zone, and interdigitation zone in the foveal area. Although there was reabsorption of the macular exudation after treatment initiation, the loss of the outer retinal layers persisted in the fovea during the follow-up period.

Retinal exudation is derived from leaking retinal vessels and composed of lipidic and proteinaceous material that settle in the outer retinal layers. When deposited within foveal area and after a period (usually months), this exudation may result in significant visual loss because of photoreceptor damage.[23] Outer retina damage because of retinal exudation has been reported in central serous chorioretinopathy, familial exudative vitreoretinopathy, and age-related macular degeneration.[24–26] In our case as in such diseases, the outer retinal damage represented by disruption and loss of the external limiting membrane, ellipsoid zone, and interdigitation zone was probably a consequence of intraretinal exudates within the outer retinal layers. We report a unique case of neuroretinitis associated with *B. quintana* and its associated SD-OCT features.

Key words: *Bartonella quintana*, neuroretinitis, spectral-domain optical coherence tomography, outer retina.

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