

Bartonellosis causing bilateral Leber neuroretinitis: a case report

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PURPOSE. *Bartonella henselae* is the causal agent of cat scratch disease and one variation in its presentation is Leber neuroretinitis. The unilateral presence of exudation as a macular star and papilledema represent its most common presentation.

METHODS. *Observational case report.* A 7-year-old girl presented a sudden decrease of visual acuity and bilateral macular exudation (macular star) as well as choroiditis.

RESULTS. A complete recovery of visual acuity was seen after a 6-week follow-up. Erythromycin plus deflazacort treatment was given.

CONCLUSIONS. The present case represents an unusual variety of cat scratch disease. This represents a challenge in the differential diagnosis of diseases such as Lyme disease and tuberculosis among others. (*Eur J Ophthalmol* 2009; 19: 307-9)

KEY WORDS. *Bartonella henselae, Neuroretinitis, Macular star*

Accepted: July 24, 2009

INTRODUCTION

Since it was identified in the early 1990s, *Bartonella henselae* (previously known as *Rochalimaea henselae*) has been associated with a wide range of eye conditions including chorioretinitis, serous macular detachment, optic nerve inflammation, vascular lesions, and Parinaud oculoglandular syndrome. Neuroretinitis or Leber idiopathic neuroretinitis is a syndrome characterized by sudden decrease in visual acuity, papilledema, lipid exudation (macular star), and usually unilaterally affecting patients of pediatric age. The differential diagnosis includes Lyme disease, leptospirosis, toxoplasmosis, tuberculosis, syphilis, and a great number of viral diseases as well as retinal vascular dysgenesis (Coats disease). Cat scratch disease's incidence has been calculated as 9.3 per 100,000, making it an uncommon disease (1). An even less common presentation is bilateral involvement, with few cases reported (2).

Case report

A 7-year-old girl presented a sudden decrease in visual acuity with an approximately 1-week evolution without any signs of malaise. She had a history of living closely with dogs; in fact, 1 week before her visual decrease one of them scratched her on the face.

Her blood pressure was 90/60 mm Hg, with a heart rate of 60 bpm and a temperature of 36.4°C.

She had no palpable preauricular or submandibular adenopathies.

She had a visual acuity of 5/400. The anterior segment was normal. In the posterior segment severe exudation with a star shape in both macular areas was seen with light hyperemia of both papillae and no blurring of its margins or tumefaction. Small choroidal lesions were seen as well (Fig. 1).

We carried out tests for antibodies against *Borrelia burgdorferi*, anti-toxoplasma, VDRL, and FTA-ABS, with negative results in all of them. Cell blood count, urea, cre-



Fig. 1 - Macular star at presentation showing choroidal lesions (arrow).

atinine, and fasting glycemia were normal. ELISA search for antibodies against *B henselae* were positive. Blood culture for *B henselae* came out negative.

Based on the clinical presentation and the anti-Bartonella antibodies, we diagnosed a severe bilateral atypical neuroretinitis or cat scratch disease. We gave treatment based on erythromycin estolate in a 250 mg qid po and deflazacort 30 mg qd po tapering it for a 6-week period. At the end of follow-up (6 weeks), visual acuity was 20/20 in both eyes with complete resolution of all retina and choroidal lesions.

DISCUSSION

Pathogenesis of neuroretinitis by *B henselae* still has not been explained thoroughly despite multiple efforts done with molecular biology techniques (3), yet it is obvious that in most cases both neural and retinal tissue are affected. Traditionally, the disease has been associated with contact with cats, yet other domestic animals (1), such as dogs or rodents, can transmit it. In fact, there has been research documenting neuroretinitis and uveitis in dogs and identifying bacteria of Bartonella gender in those cases but from different species (*vansonii* subspecies *berkhoffi*) (4) that could come out positive for cross reaction in antibody tests against *B henselae*. In the retinal findings, there can be hemorrhage, nerve fiber layer infarct (cotton wool spots), and the characteristic macular star, which can persist for months when it is as large as the one in

the present case, thus delaying the visual recovery of the patient. The lipid exudates' presence on deeper retinal layers (external plexiform layer, Henle layer) explains the shape of the so-called "macular star" because of the layout of axons at this layer's level (5).

The spectrum of the disease can also include white spots retinal syndrome, venous or arterial branch occlusion, localized vasculitis, choroidal focal infiltrations (as shown in the arrows on Fig. 1), serous retinal detachment, and subretinal peripapillary angiomas lesion (5).

Diagnosis should include confirmatory antibody detection test by ELISA against Bartonella (6) or a blood culture, which is harder to perform. There are also reports of detection by a polymerase chain reaction (7).

Diseases that may cause confusion, such as Lyme disease and its characteristic erythema migrans, tuberculosis, syphilis (the great imitators), and hypertensive retinopathy (1), as well as a central nervous system condition, must be ruled out.

It was possible to determine antibodies against Bartonella in our patient, which confirmed the diagnosis along with the medical history and therapeutic response. It is important to stress that the bilateral presentation and the severe exudation are unusual characteristics and therefore only a few cases have been reported (2).

On the other hand, many authors who have described this disease (1, 5) have stressed the optic nerve head as the main target of the condition, which is one characteristic that was not present in our case—though it is possible that the neuritis had subsided at the time the patient

came to our clinic since the symptoms had begun 1 week before.

Usually the disease is self-limited, and visual acuity improves in 4 days to 2 months (5).

As for treatment, it has been debated when to start it since many of these disorders subside in a few weeks and virtually have no sequelae in the patient's vision but, in some cases, a slight and temporary papillary pallor, defects in contrast sensitivity, and visual evoked potentials defects may persist. Curiously, electroretinogram is not affected.

Though steroids have been used before as part of treatment, their use has been controversial, especially in cases with massive exudation and a bilateral presentation as in our case (5). Use of antibiotics includes doxycycline or tetracycline as first line (1). All of these treatments must be administered in a 4- to 6-week period. On the other hand, conservative management may be recommended for mild cases. We chose erythromycin for our patient because of the well-known risks of damage to growing cartilage that tetracycline and its derivatives may cause in pediatric patients.

The present case is one of the various uncommon presentations of cat scratch disease. Clinically, the most revealing feature was the presence of a macular star. Nevertheless, its bilateral presentation as well as the absence of a visible papillitis was a diagnostic challenge, which can have many uncommon presentations besides the aforementioned. By ruling out conditions such as Lyme disease among others, and with serologic confirmation, correct diagnosis could be made.

The authors have no proprietary or financial interest in this article.

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