# **Case Report**

Taiwan J Ophthalmol 2021;11: 292-295

Access this article online



Website: www.e-tjo.org DOI: 10.4103/tjo.tjo\_29\_20

# A child with a rare presentation of ocular bartonellosis

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#### Abstract:

A 6-year-old boy was referred from the optometrist for bilateral painless blurred vision of 2 weeks duration during routine screening. Upon examination, best-corrected visual acuity was 20/200 (right eye) and 20/120 (left eye). Anterior segment examination was normal for both eyes. Funduscopy showed bilateral optic disc swelling with peripapillary exudates and diffuse retinochoroiditis involving the posterior pole. Optical coherence tomography revealed diffuse retinal thickening with intraretinal fluids and cystoid changes of central fovea. Fluorescein angiography showed bilateral hot discs with vasculitis in all quadrants and large areas of nonperfusion at peripheral retina. The patient was initially treated as presumed ocular tuberculosis (TB) based on clinical presentation and history of contact with family member having pulmonary TB. Antituberculous therapy was started and both eyes received panretinal laser photocoagulation. After 3 weeks of anti-TB treatment, serology for *Bartonella* turned out to be positive. Treatment was changed to intravenous ceftriaxone for 10 days followed by oral cotrimoxazole for 6 weeks and combined treatment with oral prednisolone. Gradual clinical improvement was seen with corresponding visual gain due to the reduction of macular edema, but residual thickening remained due to its chronicity.

#### Keywords:

Cat scratch disease, neuroretinitis, occlusive vasculitis, ocular bartonellosis, retinochoroiditis

#### Introduction

Ocular bartonellosis is the eye manifestation of a systemic illness caused by the pathogen of the genus *Bartonella*. At least a dozen species belong to this genus. *Bartonella henselae* causes cat scratch disease (CSD) and peliosis of the liver (often called bacillary peliosis). It is a Gram-negative intracellular bacterium with human transmission from the scratches of domestic or feral cats, particularly kittens. Cats can be infested with infected fleas that carry *Bartonella* bacteria. CSD occurs most frequently in children under 15.

We report a case of a child with a rare presentation of severe bilateral ocular bartonellosis with neuroretinitis, and occlusive vasculitis who was initially treated as presumed ocular tuberculosis (TB) based

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on the history of contact with pulmonary TB (PTB) patient and clinical presentation. A revised diagnosis was made when serological testing for *Bartonella* came back positive. TB is always known as a mimicker and an uncommon presentation as in this child is obviously challenging to determine a prompt diagnosis and treatment approach.

#### **Case Report**

A 6-year-old boy with no underlying illness was referred for bilateral painless blurred vision of 2 weeks duration which was incidentally picked up during routine eye screening by an optometrist. It was not associated with eye redness or discharge. He had an episode of fever about a month before the onset of the ocular symptoms which resolved with oral antibiotics. He had numerous contacts with stray cats outside the house compound but denied any injuries caused by the cats during those encounters.

**How to cite this article:** Shariffudin NS, Min TW, Adnan A, Hashim H, Teo KS. A child with a rare presentation of ocular bartonellosis. Taiwan J Ophthalmol 2021;11:292-5.

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Submission: 27-03-2020 Accepted: 22-05-2020 Published: 14-08-2020

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He also had positive contact with a TB patient who was his maternal grandfather; he was diagnosed with PTB <3 years ago and had completed anti-TB treatment. The boy had completed his immunization (including Bacille Calmette-Guerin vaccination) up to his age.

Upon general examination, the patient was of medium build, afebrile with no palpable lymph nodes. There was no evidence of any scratch or bite marks. Ocular examination revealed best-corrected visual acuity (BCVA) for right eye was 20/200 and left eye was 20/120. Pupils were equal, round, and reactive. Anterior segment examination was unremarkable. Dilated bilateral fundus examination showed bilateral optic disc swelling with dense peripapillary retinochoroiditis and the lesion extensively involved posterior pole to mid peripheral retina [Figure 1]. The retina appeared thickened and edematous. On examination, no vitritis or periphlebitis seen. Optical coherence tomography (OCT) revealed bilateral diffuse retinal thickening of the macula with intraretinal fluid and central cystoid changes [Figure 2a].

Fluorescein angiogram showed bilateral hot discs with angiographic cystoid macular edema and vasculitis in all quadrants of the retina. There were also the areas of nonperfusion at the peripheral retina in both eyes, which was more widespread in the left eye [Figure 3].

Mantoux test was negative, and chest X-ray was unremarkable. Full blood count, renal profile, and liver function tests were normal. Infective screening for syphilis, leptospirosis, melioidosis, and toxoplasmosis was all negative.

The patient was initially treated as presumed ocular TB based on clinical presentation and history of exposure to a PTB patient. In contrast, his TB workup was negative. He was referred to pediatric team for co-management of TB and was started on anti-TB medications consisting of oral isoniazid 150 mg once daily (OD), rifampicin 200 mg OD, pyrazinamide 500 mg OD, and pyridoxine 5 mg OD. Indirect laser panretinal photocoagulation to the nonperfused retina was performed under general anesthesia. In view of severe diffuse macular edema, oral prednisolone (1 mg/kg) was also instituted. He developed side effects (deranged liver function test) during the initial course of anti-TB therapy; thus, treatment was withheld by pediatrician. Incidentally, it was at this time that Bartonella serology testing by immunofluorescence assay came back positive with immunoglobulin (Ig) M titers more than 1:24 and IgG titers more than 1:128. Hence, the child was treated for ocular bartonellosis and started on intravenous ceftriaxone 375 mg twice daily (BD) (50 mg/kg/day), completed for 10 days and continued with syrup cotrimoxazole 66 mg BD (4 mg/kg) for 6 weeks and



Figure 1: Fundus photography showing right eye optic disc swelling with peripapillary exudates and peripheral chorioretinitis (a) and left eye optic disc swelling with ring-shaped peripapillary exudates and peripheral chorioretinitis (b)



Figure 2: Pretreatment optical coherence tomography of the left eye showing cystic macular edema with minimal subretinal fluid (a) and posttreatment optical coherence tomography of left eye revealing less intraretinal fluid and very minimal subretinal fluid (b)



Figure 3: Left eye fluorescein angiogram showing vasculitis and area of nonperfusion at peripheral retina

tapering dose of oral prednisolone. His blood pressure and blood sugar were normal. Visual acuity showed improvement in both eyes. Right eye BCVA was 20/80 (from 20/200) and left eye was 20/70 (from 20/120). The peripapillary retinitis and retinochoroiditis in both eyes gradually regressed [Figure 4]. On follow-up, OCT showed marked reduction of intraretinal fluid and foveal thickening [Figure 2b]. At present, the cystoid macular edema was very much reduced but not completely resolved due to its chronicity.

## Discussion

The systemic manifestation of *Bartonella henselae* infection is termed CSD. Most of the patients develop a mild



Figure 4: Fundus photography of the right eye showing markedly reduced peripapillary exudates (a) and left eye showing resolving optic disc swelling and markedly reduced peripapillary exudates at 20 weeks follow-up (b)

to moderately severe flu-like illness associated with regional lymphadenopathy. Transient skin reactions, including nonspecific maculopapular eruptions and erythema nodosum, may occur in some patients. Affected nodes are frequently tender and may even be suppurative.

Ocular involvement of CSD occurs in 5%–10% of cases and is the most common nonlymphatic organ involvement.<sup>[1]</sup> Anterior segment manifestations of *Bartonella* spp. include Parinaud oculoglandular syndrome. It can affect up to an estimated 2%–5% of symptomatic patients.<sup>[2]</sup>

There is limited prevalence data of ocular bartonellosis, especially in the Asian population. Based on a study conducted by Tan *et al.*, the most common posterior segment manifestations among Malaysians is neuroretinitis (62.5%), optic disc oedema (42.1%), anterior uveitis (31.6%) followed by vasculitis and focal choroiditis which is about 10.5% and 5.3% respectively.<sup>[3]</sup> Although CSD is very common among younger patients, ocular bartonellosis is believed to have a broader age distribution.<sup>[3]</sup> To date, there are several reported cases on posterior segment involvement among young patients. To our knowledge, our patient is the youngest reported case of severe bilateral ocular bartonellosis with neuroretinitis and occlusive vasculitis.

Table 1 summarizes published case reports of ocular bartonellosis in pediatric patients. There were two females and one male, with the youngest aged 9 years and oldest was 15 years old. Based on these cases, all presented with one common feature: neuroretinitis. Ocular presentation in our patient is much more severe and associated with occlusive vasculitis and macula edema. A prompt diagnosis of ocular TB, a known mimicker and an endemic in our country, is inevitable. A history of contact with a TB patient also led us to lean toward the diagnosis of ocular TB. In general, both ocular TB and bartonellosis may present similarly with neuroretinitis, chorioretinitis, occlusive vasculitis, and macular edema, but these features are far more common in the former rather than the latter. Therefore, the serological test is helpful in distinguishing the diagnosis of ocular bartonellosis in this atypical presentation.

Early antibiotic treatment for ocular bartonellosis seems to improve visual outcome and hasten visual recovery.<sup>[3]</sup> Treatment of CSD is dependent on age, immune status, systemic manifestations, and ocular complications.<sup>[2]</sup> Antibiotic treatment options include doxycycline, macrolides, rifampin, ciprofloxacin, ceftriaxone, and co-trimoxazole. As the patient is a Pediatric patient, an infectious disease specialist was consulted regarding the selection of antibiotic for his treatment. The mainstay of treatment consists of doxycycline 100 mg two times per day for 4-6 weeks for immunocompetent patients and up to 4 months in immunocompromised individuals. However, in children under 12 years, it is better to use erythromycin than doxycycline, which can cause teeth discoloration.<sup>[7]</sup> Doxycycline (100 mg given orally twice daily) has better intraocular and central nervous system penetration than erythromycin and therefore is often preferred in patients older than 8-12 years of age where teeth discoloration is less of a concern.<sup>[8]</sup> In pediatric age group, the standard treatment for CSD is erythromycin, but it is not suitable for ocular infection due to poor ocular penetration. Hence, the combination treatment of ceftriaxone and cotrimoxazole was used in this patient. Corticosteroids may be used in addition to antibiotic treatment to help abate the inflammatory response and to fasten recovery. In a study reported by Kodama et al. in 2003, a good outcome with antibiotics and steroids, and no relapse occurred in their patients.<sup>[9]</sup> The combination treatment with systemic steroid in this child had significantly reduced the cystoid macula edema and improved quality of vision.

## Conclusion

The most common posterior involvement in ocular bartonellosis is neuroretinitis, but it may present with uncommon features that may lead to inaccurate diagnosis. A high index of suspicion and further ancillary tests are vital in determining the diagnosis in pediatric infective uveitis cases. Oral prednisolone is effective to reduce the complication of inflammation such as cystoid macular edema and optic neuropathy. The combination treatment of antibiotic and steroid enhances the recovery and visual outcome.

#### Human subject

Consent was obtained from patient's guardian for the images and other clinical information to be reported in the journal.

#### **Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form, the legal guardian

Table 1: Published case reports of ocular bartonellosis in	pediatric	patients	(2005-2019)
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Author/years	Age/gender	Visual acuity on presentation	Ocular diagnosis	Treatment	Final visual acuity
Raihan <i>et al.</i> 2014 <sup>[4]</sup>	15/male	OD: 20/20 OS: 20/300	OU: Nongranulomatous panuveitis with neuroretinitis	Oral azithromycin daily for 6 weeks and oral prednisolone	OU: 20/20
Mirri <i>et al</i> . 2019 <sup>[5]</sup>	10/female	Not mentioned	OU: Optic neuritis	IV cefotaxime	Not mentioned
Martínez-Osorio <i>et al.</i> 2005 <sup>[6]</sup>	9/female	OD: 20/25	OD: Recurrent iridocyclitis with neuroretinitis	Oral trimethoprim- sulfamethoxazole BD for 6 weeks and topical fluorometholone	Not mentioned

IV: Intravenous OU: Oculus uterque (both eyes), OD: Oculus dexter (right eye), OS: Oculus sinister (left eye), BD: Twice daily

has given his consent for images and other clinical information to be reported in the journal. The guardian understands that name and initials will not be published, and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

#### **Financial support and sponsorship** Nil.

#### **Conflicts of interest**

The authors declare that there are no conflicts of interests of this paper.

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