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Bilateral neuroretinits and exudative retinal detachment with multifocal subretinal deposits secondary to Bartonella henselae infection

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ABSTRACT

Purpose: to describe a case of bilateral neuroretinitis with bullous retinal detachment and multiple subretinal lesions, in a 10-year-old immunocompetent girl.

Observations: A broad workup for infectious, inflammatory and masquerade etiologies was done for the patient, resulting in positive IgM and IgG for Bartonella henselae. The patient demonstrated improvement in the visual acuity, and rapid resolution of the retinal detachment and subretinal lesions in both eyes in response to systemic rifampin, doxycycline and corticosteroids.

Conclusions and Importance: Bartonella henselae neuroretinitis may present as an acute form of bullous retinal detachment with multiple subretinal lesions and markedly reduced vision. Significant visual improvement may occur with prompt treatment with a combination of systemic antibiotics and corticosteroids.

1. Introduction

Cat-scratch disease (CSD) is generally a self-limiting disease caused by Bartonella henselae, a gram-negative bacillus. The clinical presentation of the disease includes regional lymphadenopathy, malaise and fever, and splenomegaly which usually follow a cat scratch or a bite.[1] Rare manifestations of secondary CSD may include osteomyelitis, endocarditis, encephalopathy, and hepatosplenic abscesses.[2–4] The ocular manifestations of CSD are uncommon and occur in 5–10% of patients with cat scratch disease.[5] The different presentations include Parinaud oculoglandular syndrome, neuroretinitis, retinochoroiditis, endophthalmitis, vascular occlusions, serous macular detachments, multiple mass-like lesions resembling ocular metastases, and retinal vasoproliferative lesions.[6–9] We report a case of bilateral neuroretinitis with a serous bullous retinal detachment extending to the inferior periphery with multiple subretinal lesions associated with cat scratch disease in a 10-year-old female. To the best of our knowledge this case of exudative retinal detachment with multiple subretinal lesions has not been reported previously.

2. Case report

A 10 year-old Caucasian female with no significant past medical history presented to the emergency department with a one-month history of headache, 1 week of right eye exotropia, and 3 days of right vision loss. An optometric exam was noteworthy for possible papilledema. The remainder of the past medical history was unremarkable.

On examination in the emergency department the patient had right exotropia, intact extraocular movements, and no light perception in her right eye. The patient was afebrile with a blood pressure of 115/74. The patient had one enlarged mandibular lymphnode but no other lymphadenopathy.

Urgent ophthalmology consultation was requested. On examination, visual acuity was no light perception (NLP) OD and 20/20 OS. Anterior segment exam revealed 2+ cell OD. Dilated ophthalmoscopic exam of the right eye demonstrated grade 4 disc edema, a macula with few well-circumscribed whitish lesions, venous tortuosity and multiple creamy white subretinal lesions with an inferior bullous retinal detachment. The left eye had grade 3 disc edema but no other findings (Figs. 2 and 3).

The patient was residing in Kentucky. She had two pet dogs but no history of travel. She also reported a recent self-resolved insect bite lateral to the right eye and remote contact with kittens that involved a

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scratch.

Urgent magnetic resonance imaging (MRI)/magnetic resonance venography (MRV) was completed which demonstrated a plaque like lesion lining the posterior right globe and covering optic disc (Fig. 1). There was no intracranial hemorrhage or mass. Brain MRV was normal. The differential diagnosis for the intraorbital lesion included retinoblastoma, melanoma or ocular toxocariasis. Broad testing for infectious, inflammatory and malignant etiologies was initiated.

The patient underwent a lumbar puncture; the opening pressure was measured at 23 cm of water. Fluid was clear with 60 mg/dl glucose, 1 WBC and 16 mg/dl protein. Gram stain was negative for bacteria and cytology was negative for malignant cells.

Two days after original presentation, macular exudates appeared in the left eye. Results for Syphilis, HIV, Proteinase, MPO (Myeloperoxidase), Quantiferon, ACE, Lysosome, and EBV IgM were negative, while EBNA (Epstein Barr Nuclear Antigen) was positive. Due to high suspicion for neuroretinitis the patient was put on empiric treatment with doxycycline and rifampin to cover for Bartonella henselae.

Five days after presentation and three days after initiation of systemic treatment, the vision improved to light perception (LP) in the right eye. Tests for Histoplasma, Tuleremia, Toxoplasma, Toxocara, Anaplasma *phagocytophilum*, Pan-Ehrlichia, Rickettsia *rickettsii*, Borrelia *burgdorferi* IgG/IGM were negative but positive for Bartonella henselae IgM and IgG (titer of 1:16 and 1:1024 respectively). CSF pathogen panel was negative for multiple organisms, including Streptococcus *pneumonia,* Listeria *monocytogens*, Herpesviruses, Cytomegalovirus, and Cryptococcus species. Treatment consisted of systemic antibiotics for four weeks, as well as oral prednisone. Over the next month, there was resolution of bilateral disc edema, exudates, exudative retinal detachment, and macular edema. At six months vision had improved to 20/160 OD and remained 20/20 OS, with optic nerve pallor OD (Figs. 2 and 3). The patient remained symptom free during her follow up of 8 months, 7 months off of therapy.

3. Discussion

This child had an atypical presentation of NLP vision and extensive exudative retinal detachment with multiple subretinal lesions in the right eye. She ultimately recovered vision to 20/160, but sustained optic nerve damage and outer retinal layer loss on optical coherence tomography.

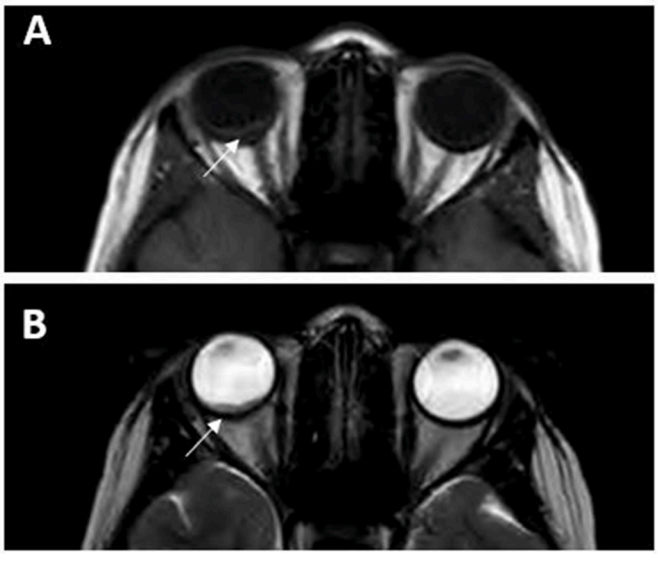

Fig. 1. Contrast-enhanced MRI demonstrating a plaque-like lesion lining the posterior pole of the right globe correlating with the exudative retinal detachment, which was hyperintense in T1-weighted image A and hypointense in T2-weighted image B (white arrow).

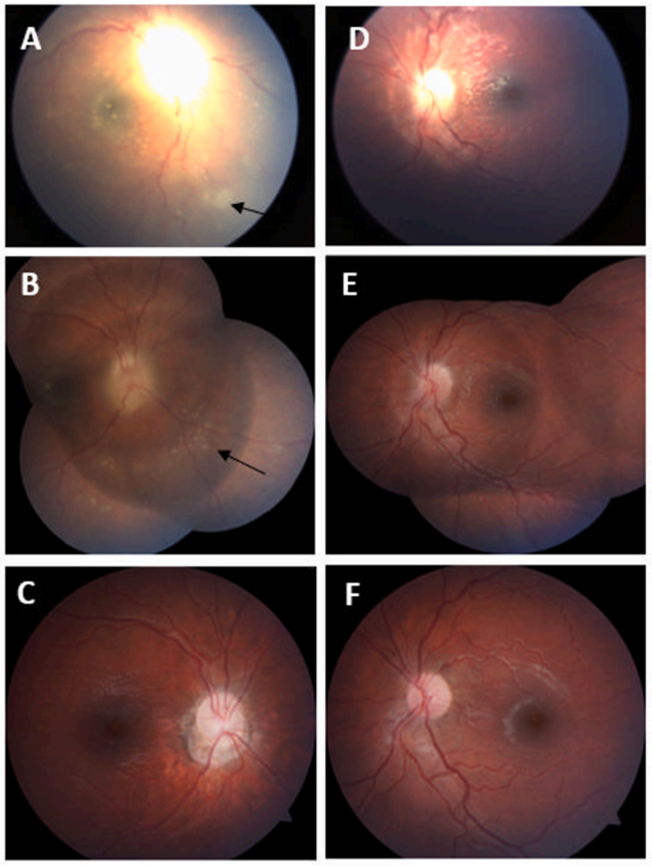

Fig. 2. Right eye with grade 4 disc edema with tortuous vasculature and inferior subretinal exudation with multiple subretinal lesions (black arrows) (image A), gradually improved over two weeks and 6 months (image B and C respectively) However, optic nerve pallor resulted OD. Left eye with grade 3 disc edema with incomplete macular star (image D), gradually improved over 2 two weeks and 6 months (image E, and F respectively).

Five to ten percent of patients with CSD may develop ocular pathology.[5] In this case there were no systemic symptoms apart from an enlarged lymph node of the neck, headache, and fatigue. In spite of its low incidence, the finding of regional lymphadenopathy/lymphadenitis on physical examination may be an important clue to the diagnosis of CSD and lymph node biopsy may allow for diagnosis by polymerase chain reaction. Ocular manifestations of CSD include optic nerve lesions, neuroretinitis, uveitis, retinitis, chorioretinitis, endophthalmitis, vascular occlusions, multiple mass-like lesions resembling ocular metastases, retinal vasoproliferative lesions and serous macular detachments.[6–9] About 20% of ocular CSD are bilateral[6] and 2% of all patients present with bilateral neuroretinitis.[6,7,10] Bartonella-related bilateral neuroretinitis is rare in children, with few case reports in the literature.[11–14] The unique feature in this case is the large bullous serous retinal detachment that extended to the periphery with multiple subretinal deposits, making the initial diagnosis more difficult. Submacular serous retinal detachment has been described in CSD but is usually limited to the posterior pole.[15–17] Toshihiko et al. reported retinal detachment secondary to Bartonella henslae endophthalmitis,[18] and Ulrich et al. described a case with exudative retinal detachment in a child, but there was no evidence of multiple subretinal lesions.[19]

As most patients with CSD have a self-limited course, the value of antibiotic therapy for CSD as well as other treatment modalities is debated.[8,9,20–22] Antibiotic treatment is often considered in severe systemic disease, immunocompromised patients, or sight-threatening ocular involvement, including neuroretinitis.[8] Multiple antibiotic

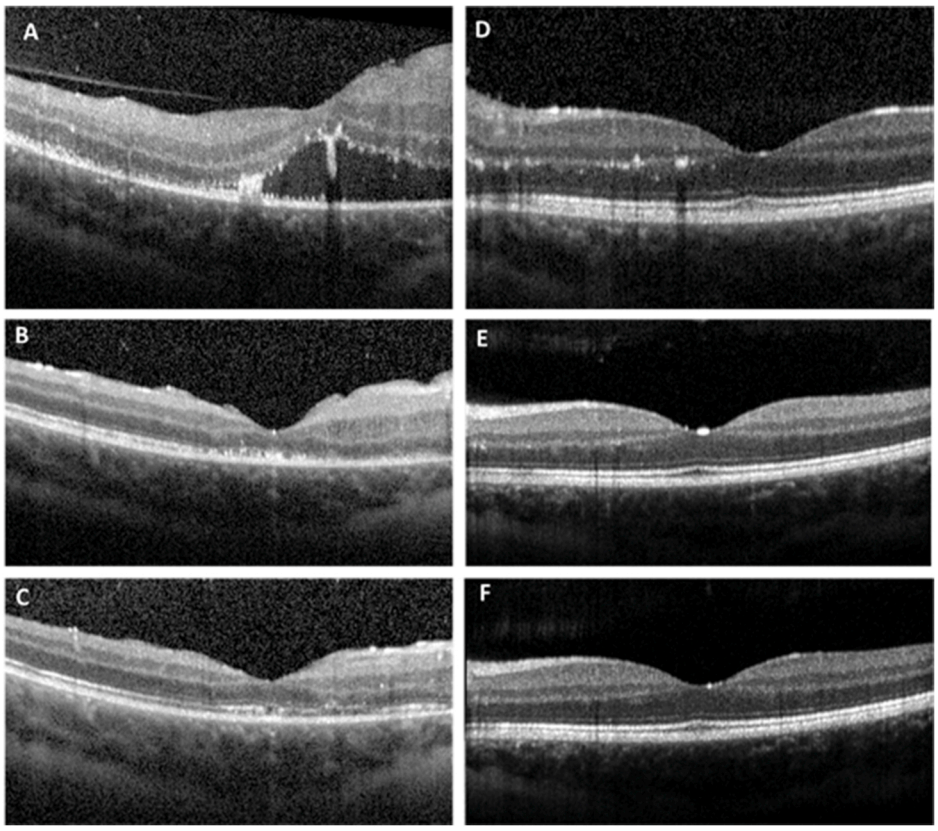

Fig. 3. OCT image of right eye at 2-week clinic follow-up showing subretinal fluid with shaggy photoreceptors (A) which resolved at 2-month follow-up but with atrophy of the ellipsoid zone (B) At six-month follow-up partial recovery of the ellipsoid zone is seen (C). The left eye showed hyperreflective foci at the outer nuclear layer (ONL) at 2-week follow-up (D) with partial resolution at 2-month follow-up (E), and complete resolution at six-month follow up (F).

combinations have been used to treat CSD.[7,9] In this case the combination of doxycycline and rifampin prescribed because of excellent CNS penetration.[7] A recent retrospective study by Wilner et al. concluded that the use of oral corticosteroids in combination with antibiotics improves the visual outcome in patients with CSD neuroretinitis.[6] However, a review by Chi et. Al. concluded that antibiotics were ineffective.[20] In our patient, the large exudative retinal detachment resolved relatively quickly and the subretinal lesions began to fade shortly after initiation of treatment. The patient improved significantly with a combination of corticosteroids, doxycycline, and rifampin. However, visual acuity did not recover completely in the right eye, with optic atrophy and damage to the ellipsoid layers on OCT (Fig. 3).

4. Conclusion

Bartonella henselae neuroretinitis can uniquely present as a bullous retinal detachment with multiple subretinal deposits. Significant visual improvement may occur with prompt treatment with a combination of systemic antibiotics and corticosteroids.

- Written consent to publish this case has not been obtained. This report does not contain any personal identifying information.

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None.

Declaration of competing interest

None.

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