

# Optic neuritis and chorioretinitis as ocular manifestations of borreliosis in Brazil: three cases reported

## *Neurite óptica e coriorretinite como manifestações oculares da borreliose no Brasil: três casos relatados*

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### ABSTRACT

*Lyme disease is a systemic infection caused by a tick bite and transmission of the Borrelia burgdorferi spirochete. Species of tick vectors of the disease infest mainly wild or rural animals and rodents that may be asymptomatic reservoirs of the bacteria. Characteristic of the northern hemisphere, Lyme disease in Brazil takes on different characteristics, complicating diagnosis. This paper aims to describe three cases of Lyme-like disease in a city in the state of Bahia, Brazil, with ophthalmologic findings.*

**Keywords:** Lyme Disease, ticks, Lyme-like Disease, Borrelia burgdorferi

### RESUMO

A doença de Lyme é uma infecção sistêmica causada pela picada do carrapato e transmissão da espiroqueta Borrelia burgdorferi. As espécies de carrapatos vetores da doença infestam, principalmente, animais silvestres, rurais e roedores que podem ser reservatórios assintomáticos da bactéria. Característica do hemisfério norte, a doença de Lyme no Brasil assume características distintas, dificultando seu diagnóstico. Esse trabalho tem por objetivo, descrever três casos da doença Lyme símile do Brasil, com achados oftalmológicos, em município do Estado da Bahia.

**Descritores:** Doença de Lyme, carrapatos, Doença Lyme símile, Borrelia burgdorferi

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## INTRODUCTION

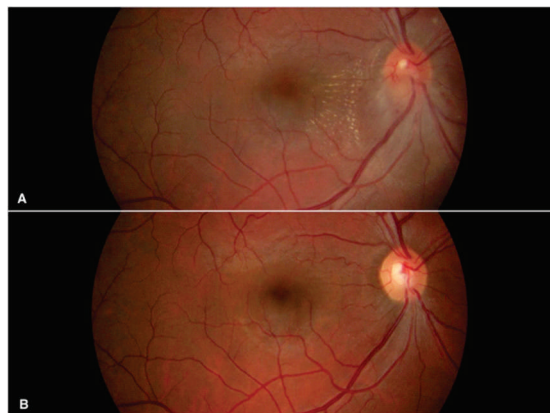
**L**yme disease (LD) is an exotic zoonosis from the northern hemisphere, caused by the bite of the tick *Ixodes Castor* and transmission of *Borrelia burgdorferi sensu lato*. In Brazil, *Borrelia burgdorferi* has never been isolated, and this tick species is not found in endemic areas. The clinical, epidemiological and laboratory characteristics of the infectious disease found in this country diverge substantially from those classically reported in patients with Lyme disease elsewhere in the world, consequently receiving the denomination Baggio-Yoshinari Syndrome (BYS).<sup>(1)</sup> As in LD, the Brazilian Human borreliosis may present cutaneous manifestations, as well as osteo-articular, neurologic, cardiac, psychiatric, ocular and immune-allergic.<sup>(2,3)</sup> The different clinical aspect is the high number of relapses, especially if the patient is not diagnosed and treated early. The tick species probably involved in Brazil's disease are *Cajennense amblyomma* and *Rhipicephalus microplus* and the infectious agents, latent spirochetes in cystic form, very different from the spiraled ones found in the northern hemisphere. It is possible that the geographical conditions and biodiversity in Brazil have generated a mutant bacterium originating with complex *B. burgdorferi* responsible for the production of low and oscillating patient's antibody.<sup>(4)</sup> This fact, together with flu-like clinical features, common to so many other infectious syndromes, hinders very much the identification of cases.<sup>(1)</sup> The objective of this study is to report three cases of ocular borreliosis in the city of Irecê, Bahia, Brazil, diagnosed based on epidemiological, clinical, eye fundus findings and serology.

## CASE REPORT

### Case 1

Male 35 years old, was raising a fox at his residence. Febrile syndrome for 03 weeks with low vision in the right eye (RE) during this period, no complaints in left eye (LE). Weight loss. Best corrected visual acuity (BCVA) in RE 20/80, and LE 20/20. Anterior segment and intraocular pressure was normal. Eye fundus exam: RE = optic neuritis, retinitis, macular star forming, serous detachment of the retina below the disk, retinal hemorrhages, cotton wool spots. (A) LE = Cotton wool spot near the inferior nasal vessels. Serology for *B. burgdorferi*, indefinite, and negative for toxoplasma, toxocara, bartonella and syphilis.

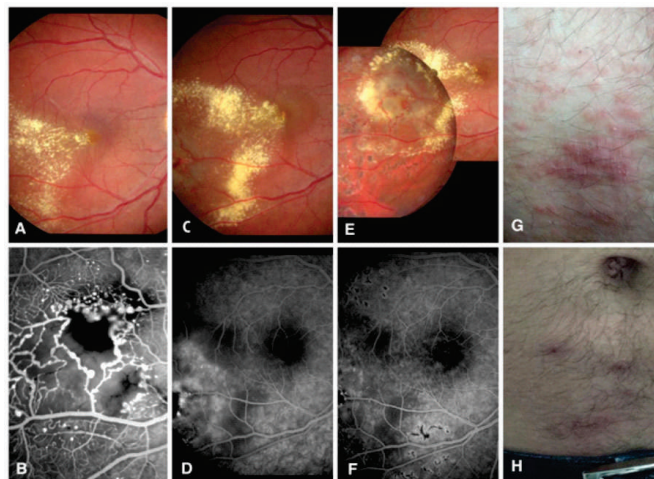
Treatment: doxycycline 100 mg for 04 weeks, combined with prednisone, with satisfactory results. (B) BCVA at end: 20/20. (Figure 1)



**Figure 1:** A : Eye fundus before treatment B : Eye fundus after treatment.

### Case 2

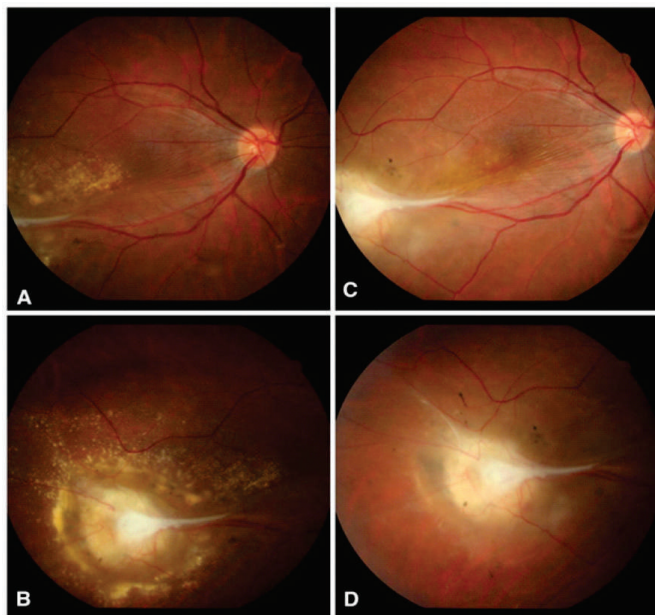
Male 32 years old. Electrician, used to do installations in rodent-infested warehouses in the countryside. Febrile syndrome for 06 weeks, which was referred to as prolonged flu and discouragement for daily activities. Low vision RE for 10 days. BCVA 20/200 RE and 20/20 in the LE. Anterior segment and intraocular pressure was normal. Eye fundus exam: RE = nodular lesion of the inferior temporal retina, causing vasculitis, saccular dilatation of the vessels, ischemic area distal to injury, exudation, and serous retinal detachment. Coats-like disease. ( A,B ) Serology for *B. burgdorferi* (Enzyme-Linked Immunosorbent Assay ELISA), IgM positive. Negative serology for syphilis, HIV, toxoplasma and Bartonella. Tuberculin test and chest X-ray normal. Western blotting collected during treatment, detected one band, being considered negative. Presence of placoid erythema in abdomen. Treatment: Laser in the ischemic area. Patient progressed with worsening of exudation. (C,D) After completion of laboratory tests doxycycline 100 mg was introduced for 04 weeks, combined with prednisone, with complete resolution of the retinal detachment. Skin injury healed. ( E,F) BCVA at end: 20/25. (Figure 2)



**Figure 2:** A, B : Fundus findings at the first visit. C, D : Worsening of exudation after laser. E, F : Complete resolution of the retinal detachment after doxycycline. G, H : Skin injury healed

### Case 3

Female, 27, spent vacations on a farm, where she had contact with horses and cattle. Complaints of low vision in both eyes, for 02 years, with episodes of worsening during this period. Empirically treated for recurrent retinal lesions, considered as toxoplasmosis in another service. Complaints of poor appetite and chronic fatigue. BCVA 20/100 RE and 20/40 in the LE. Anterior segment and intraocular pressure was normal. Eye fundus exam: RE = nodular lesion in inferior temporal retina, with surrounding exudation. Epiretinal membrane formation, from the lesion to the temporal vessels. ( A,B ) LE = nodular lesion in the macular area, healed, without signs of inflammatory activity in the first evaluation. Serology for *B. burgdorferi*, positive, and negative for Bartonella, toxocara, syphilis and toxoplasma. Antinuclear Antibody negative, angiotensin converting enzyme normal, tuberculin skin test and chest X-ray normal. Treatment: doxycycline 100 mg, for 04 weeks, combined with prednisone, with improvement of inflammation. ( C,D ) BCVA at end: 20/100. (Figure 3)



**Figure 3:** A, B : Fundus findings at the first visit. C,D Eye fundus after treatment.

## DISCUSSION

Human borreliosis in Brazil has some particular characteristics. Geographical conditions and the country's biodiversity predispose it to the development of atypical spirochetes adapted to live in vertebrate and invertebrate hosts. These borrelia with atypical morphology tend to live within the host's cells, causing increased resistance to antibiotics and serum levels below those considered for Lyme disease criteria in the world.<sup>(1,5)</sup>

For all that, it was made necessary the establishment of criteria to assist in the identification of Brazilian cases, with epidemiology, clinical signs (joint, eye, cardiac and neurological) and positive serology, being the major criteria. Methods for serological evaluation are the enzyme-linked immunosorbent assay (ELISA) and Western blotting, and the antigen used in the absence of a Brazilian isolate is of American origin. The interpretation of these tests should be done carefully, given the possibility of cross-reactivity, false positive and false negative, as may occur, for example, in patients who have received treatment.<sup>(6)</sup> The minor criteria are chronic fatigue syndrome, visualization of spirochetes in the dark field and recurrences. The diagnosis is confirmed in the presence of three major parameters or two larger and two smaller.<sup>(1)</sup>

At the site of the bite, there may be a rash that progresses in centrifugal direction, and classically evolves to form rings, there may be variations such as triangular or placoid aspect.<sup>(6)</sup>

Ocular symptoms in classic Lyme disease can range from conjunctival hyperemia and keratitis, to intermediate uveitis, optic neuritis, cotton wool spots, vascular occlusions or inflammation at the level of the choroid and retina that occur with vitreoretinal proliferation.<sup>(7-9)</sup> In the Lyme-like disease in Brazil, there are few reports of ocular involvement, making the documentation of new cases of great importance for a better understanding of the disease.<sup>(2-10)</sup>

Recurrence may occur in up to 75% of cases that are diagnosed in late stages (more than three months from the onset of the disease) and ocular symptoms may occur in up to 35% of patients.<sup>(1)</sup>

Treatment of BYS is done with doxycycline 100 mg twice daily for 4 weeks. For patients diagnosed at late stage, treatment should be extended for 3 months and those with neurological symptoms should receive ceftriaxone 2g intravenously and after that, should follow with use of doxycycline, twice daily for three months.<sup>(4)</sup>

Patients with inflammatory retinal lesions should be investigated for borreliosis. Especially those who have visited endemic areas or who have had contact with ticks. Diagnosis requires more attention, since the disease can present very similar symptoms to other infectious syndromes and some patients may develop to chronic phase, when it becomes even more difficult the correlation between some findings and the initial epidemiological history. Also, because it is an infectious and contagious disease, screening of endemic areas becomes essential for adequate control of risk situations.

## REFERENCES

1. Yoshinari NH, Mantovani E, Bonoldi VL, Marangoni RG, Gauditano G. Doença Lyme-símile brasileira ou síndrome baggio-Yoshinari: zoonose exótica e emergente transmitida por carrapatos. *Rev Assoc Med Bras.* 2010; 56(3): 363-9.
2. Sato MT, Schmitt A, Greboge P, Arana J, Moreira AT, Yoshinari NH. Neurorretinite associada à ceratite intersticial: relato do primeiro caso de doença de Lyme no Estado do Paraná. *Rev Bras Oftalmol.* 2003;62(4):275-83.
3. Yoshinari NH, Barros PJ, Bonoldi VL. Perfil da borreliose de Lyme no Brasil. *Rev Hosp Clin Fac Med São Paulo.* 1997;52(2):111-7.
4. Gouveia EA, Alves EF, Mantovani E, Oyafuso LK, Bonoldi VL, Yoshinari NH. Profile of patients with Baggio-Yoshinari Syndrome admitted at "Instituto de Infectologia Emilio Ribas". *Rev Inst Med Trop S Paulo.* 2010;52(6):297-303.
5. Mantovani E, Costa IP, Gauditano G, Bonoldi VL, Higuchi ML, Yoshinari NH. Description of Lyme disease-like syndrome in Brazil. Is it a new tick borne disease or Lyme disease variation? *Braz J Med Biol Res.* 2007;40(4):443-56. Review.
6. Fonseca AH, Salles RS, Salles SA, Madureira RC, Yoshinari NH. Borreliose de Lyme símile: uma doença emergente e relevante para a dermatologia no Brasil. *An Bras Dermatol.* 2005;80(2):171-8.
7. Jacobson DM. Lyme disease and optic neuritis: long-term follow-up of seropositive patients. *Neurology.* 2003;60(5):881-2.
8. Klaeger AJ, Herbort CP. Cotton wool spots as possible indicators of retinal vascular pathology in ocular Lyme borreliosis. *Int Ophthalmol.* 2010;30(5):599-602.
9. Mikkila HO, Seppala IJT, Viljanen MK, Peltomaa MP, Karma A. The expanding clinical spectrum of ocular Lyme borreliosis. *Ophthalmology.* 2000;107(3):581-7.
10. Fugimoto F, Ghanem RC, Monteiro ML. Pupila tônica bilateral como seqüela oftálmica isolada da doença de Lyme: relato de caso. *Arq Bras Oftalmol.* 2005;68(3):381-4.

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