

Case Report

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Diplopia as the Sole Symptom of Lyme Borreliosis

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ABSTRACT

Lyme Borreliosis (LB) is the most common human tick-borne disease in the Northern Hemisphere.

The illness usually begins with erythema migrans, followed by neurologic or cardiac abnormalities and finally arthritis. Ocular complications have been reported, including conjunctivitis, keratitis, uveitis, oculomotor palsies, papilloedema and others, but they have been rarely described in literature as unique clinical feature of disease.

Herein, the case of a 53-year-old Caucasian woman with diplopia as the sole symptom of LB. No other systemic symptoms were detected, leading to an initial misdiagnosis.

Undergoing to detailed investigation, our patient actually had serology positive for Borreliosis.

Two months after antibiotic and orthoptic treatment, diplopia definitively disappeared.

These results confirm the theory that an appropriate drug therapy associated with an effective orthoptic rehabilitation has made possible a quick restoration of binocular vision, achieving a good quality of life.

This case demonstrates the importance of considering LB as part of the differential diagnosis of patients with isolated cranial nerve palsies, stressing the importance of the orthoptic intervention.

KEYWORDS

Abducens Nerve Palsy; Diplopia; Eye Movement Training; Lyme Borreliosis; Orthoptic Evaluation; Prism Rehabilitation.

INTRODUCTION

Lyme Borreliosis (LB) is the most frequently infectious multi system disorder worldwide [1].

The prevalence of LB is estimated to be 20-100 cases per 100,000 persons in the United States and 100-155 cases per 100,000 persons in Europe [2].

The disease is transmitted following exposure to a tick bite carrying the spirochete Borrelia Burgdorferi [1, 3].

It typically begins with an erythematous rash (erythema migrans), but patients can develop also cardiac anomalies (4-8%), neurologic compromise (11%) and arthritis (45-60%) [3].

Literature also reports common ocular manifestation, such as conjunctivitis, keratitis, uveitis, oculomotor palsies, papilloedema, papillitis, accompanied by ophthalmological symptoms among pain, visual impairment, photophobia, myodesopsia, diplopia and lack of accommodation [4-5]. LB diagnosis is highly likely based upon appropriate serology and clinical manifestations.

Symptoms generally disappear with antibiotic treatment that generally lasts less than 4 weeks, although the length of therapy could vary according to disease course [1-3].

Ocular LB is probably underdiagnosed due to a difficult serologic diagnosis, as well as its various and soft ophthalmological symptoms.

This case presents a 53-year-old Caucasian woman affected by an isolated paralytic strabismus originally caused by LB.

The clinical findings, limitated to diplopia, have lead to a late diagnosis since an isolated abducens nerve palsy does not represent a specific features of Borreliosis.

CASE REPORT

A 53-year-old Caucasian woman presented to the Orthoptic

and Ophthalmologic Assistance Unit of Careggi Hospital with a 1-month history of diplopia.

She was admitted few days before to the Department of Internal Medicine for further clinical investigations.

She had no fever, headache, orbital pain and any other symptoms. Her medical history was negative for recent trauma and systemic diseases. She denied neurological or thyroid diseases, diabetes or other vascular affections.

She reported to live in a countryside area near Florence, working as a farmer in her agricultural property.

On examination, she had a best corrected visual acuity of 20/20 in both eyes using her own spectacles: right eye +1.00 DS and left eye +1.75 DS.

Pupils were round, equal and reactive to light, with no relative afferent pupil defect.

Corneal reflections showed a primary position left esotropia (ET) larger for distance than near. Diplopia was horizontal, with an intermittent oscillopsia sensation in lateral gaze.

Prism cover test (PCT) revealed 25Δ ET at distance and 10Δ ET' at near. Extra Ocular Motility (EOM) revealed limited abduction on left gaze associated to a mild left-beating nystagmus. The Hess chart showed underaction of the left lateral rectus (LR), with the development of muscular sequelae according to Hering's law.

Waiting for the clinical diagnosis of Internists, she was advised on a penalization therapy using an opaque foil over the left eye to avoid diplopia. The patient was also trained with eye movement exercises to strengthen the left LR, facilitating the restoration of single vision.

A month later, she came back again for her orthoptic followup. Meanwhile, the diagnosis of LB was established with specific serology test requested thanks to a medical intuition: on closer questioning, the patient reported the possibility of having been bitten by a tick carrying an infective bacteria during her work in the countryside.

Doxycycline (100 mg twice a day for three weeks) was administered and the second orthoptic evaluation showed a good improvement in left abduction, with regression of diplopia.

Given the smaller strabismus angle for distance, a temporary prism correction to restore binocularity and align the visual axes was proposed.

A 10 Δ base-out (BO) prism was the minimum correction required to re-establish binocular single vision (BSV). This was prescribed as a 10 Δ BO temporary Fresnel prism, pending further improvement of her condition.

She was encouraged to insist with eye movement training. Two months after completing her therapy and orthoptic treatment, the patient experienced a complete clinical resolution of diplopia without prism correction lenses, recovering her pre-existing BVS and stereopsis.

DISCUSSION

Lyme Borreliosis, a multi-system organ disorder caused by Borrelia burgdorferi, is the most common human tick-borne disease in the Northern Hemisphere [1-3].

This complex syndrome can be highly difficult to diagnose, not only for its disparate symptoms, but especially because people often do not consider to have been exposed to ticks.

In United States approximately 25-30% of patient with early LB recall the tick bite (in Europe 64%), so clinicians must direct the history toward this possibility [6].

LB can be divided into three clinical stages: early localized, early disseminated and late disseminated.

Stage I is generally characterized by erythema migrans, that occurs at the site of the tick bite.

Stage II begins weeks or several months after infection, with multiorgan involvement of cardiovascular and central nervous system.

Stage III, the chronic phase, occurs months after the initial onset, affecting the joints, peripheral nervous system and subcutaneous tissues [1-3].

Ocular involvement has been reported during all stages of LB [4-5, 7-11].

In fact the spirochete may also remain dormant in the eye, accounting for late ocular manifestations [5, 11].

Conjunctivitis, photophobia and periorbital edema have been described during early phase.

In addition, during early as well as and late disseminated LB, keratitis, iridocyclitis, vitritis, multifocal choroiditis, exudative retinal detachment and panophthalmitis have been reported [4-5, 9-11].

Moreover, neuro-ophthalmologic manifestations including optic neuritis, disc edema and oculomotor palsies may be also observed [5, 7-10, 13-14].

Isolated oculomotor weakness is due to III, IV, or VI cranial nerve palsies, which may occur individually or in combination with other neurological abnormalities. Diplopia is the typical ocular complaint in these instances [10-11, 13-14].

Most described cases of diplopia associated with LB are secondary to abducens palsy [5, 7, 10, 12-13].

Because of its nonspecific ocular symptomatology, a patient with isolated VI nerve palsy without a sure diagnosis should be questioned for exposure to an endemic area for LB, previous tick bites, skin rash, or arthritis [12-15]. In these cases, differential diagnosis must include Borreliosis, multiple sclerosis, viral infection, syphilis, sarcoidosis, and vascular diseases [15].

The prognosis for patients with ocular LB is generally excellent when they are treated early with appropriate antibiotic regimens [10].

Antibiotic selection and duration of therapy are chosen based on the patient's clinical manifestations and stage of disease [3, 11].

There are no evidences that extended treatment changes its natural history, but surely an early treatment can reduce the risk of chronic disease [1, 3].

Current treatment recommendations for neurologic early disseminated LB, including meningitis, cranial nerve palsy, or radiculitis, suggest principally doxycycline, amoxicillin, or ce-furoxime axetil [3, 11, 16].

Isolated cranial nerve palsy is usually treated with oral administration at least for 14-21 days [16-17].

Like other neuropathies, oculomotor nerves palsies generally resolve within two weeks to five months after onset with appropriate treatment [11].

CONCLUSIONS

Ocular LB is probably underdiagnosed not only because of difficulties in the serologic diagnosis, but also because its clinical ocular features are various and nonspecific.

Although many ocular manifestations have been described in literature, an isolated abducens palsy as the sole clinical symptom of LB has been rarely reported. [5, 7, 10, 12-13]

In endemic areas, people with unexplained, acquired ocular motor abnormalities should be always evaluated for Borreliosis infection. [5]

This report shows a 53-years old patient affected by monolateral abducens palsy with diplopia.

Some time before, she had been bitten by a tick, probably without realizing it at the moment, but developing Borreliosis as consequence.

Thanks to detailed medical history, serologic and clinical tests, her paralytic strabismus had been classified as a rare feature of LB.

Antibiotic therapy, associated with orthoptic rehabilitation treatment, solved completely her condition.

This case should increase not only consciousness of clinicians to include LB in differential diagnosis for unexplained cases of diplopia, but also that a careful orthoptic evaluation helps to get a prompt diagnosis and a quick recovery.

In fact, timely assessments and appropriate rehabilitative interventions - such as eye movement training and prism therapy - surely concur to improve acquired oculomotor nerve

palsies.

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