

# Paralytic Strabismus as a Manifestation of Lyme Borreliosis

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## ABSTRACT

*Lyme disease is a multi-system organ disorder caused by Borrelia burgdorferi. Although ocular manifestations have been reported, these remain a rare feature of the disease. This report shows a 49-years old patient that has been bitten by a tick and as consequence of which developed symptoms of the Lyme disease. In 1998 the patient was hospitalized in our Eye Clinic due to operating treatment of the paralytic strabismus (abductal nerve paralysis), as a rare feature of the Lyme disease. Postoperative squint angle was significantly reduced, but without any temporal movement. Diplopia was still present, though slightly reduced with the use of prism eyeglasses. The improvement of the quality of life was achieved, as well as the patient's satisfaction.*

**Key words:** Lyme disease, paralytic strabismus, therapy

## Introduction

Lyme borreliosis (Lyme disease) is multisystem disorder which can affect a complex range of tissues including the skin, heart, nervous system, and to a lesser extent the eyes, kidneys and liver. The illness is caused by a spirochete (spiral-shaped bacteria) *Borrelia burgdorferi*, which is transmitted during the blood feeding of ticks of the genus *Ixodes*<sup>1–3</sup>. It is one of the most frequent zoonosis in Croatia, apart from salmonellas. As an illness itself, Lyme disease was described in 1977, in the town of Old Lyme, Connecticut, USA, after a mysterious outbreak of arthritis.<sup>4</sup> *B. burgdorferi* is a slow growing, fastidious organism which requires a complex liquid medium and an optimal temperature of 33–35 °C for growth. A large number of ticks have been reported as carries of *B. burgdorferi*. In Europe, the main vector is *Ixodes ricinus*. The process of transmission occurs through salivation during the feeding process on an animal host. For transmission of spirochetes to cause an infection, at least 24 hours are needed. The infection is usually acquired from a reservoir host that are mostly small and medium sized animals, birds, or also larger animals such as sheep, deer, cows or horses. Infection can be subclinical (asymptomatic) or can have various clinical manifestations, depending on the affected tissue, the duration of the infection, host factors such as vulnerability of the immune system and immunogenetic

factors which could predispose to the development of certain complications<sup>3–5</sup>. Clinical presentation can generally be divided into three stages (progression from an early to later stage is not inevitable, especially when the infection is untreated):

### 1. Early localised Lyme borreliosis

In approximately 60% of cases the early clinical feature of LB is erythema migrans, the characteristic red rash or lesion spreading from the site of a tick bite. The affected patient may also have vague »flu-like« symptoms<sup>3,5</sup>.

### 2. Early disseminated Lyme borreliosis

The organism may spread to other tissues via the bloodstream and lymphatic, and activity can continue for over a year after the initial infection. Manifestations of this stage may include more severe »flu-like« illness, cranial nerve lesions, aseptic (»viral-type«) meningitis, encephalitis, arthritis, carditis<sup>3,5</sup>. Ocular manifestations of this stage of Lyme borreliosis are follicular conjunctivitis, keratitis, scleritis, uveitis, haemorrhages, optic neuritis, orbital myositis and eye muscle paresis<sup>3,4,6–11</sup>.

### 3. Late Lyme borreliosis

Progression to this stage is uncommon but may occur in patients who were not treated or inadequately treated at an earlier stage. The most frequent of them are: chronic Lyme arthritis, acrodermatitis chronica atrophicans, and chronic Lyme meningoencephalitis with peripheral nervous system complications<sup>3,5,6</sup>. Diagnosis of Lyme disease should take into account: history of possible exposure to ticks, especially in areas where Lyme disease is known to occur; symptoms and signs; the results of blood tests used to determine whether the patient has antibodies to Lyme disease bacteria<sup>5</sup>.

Lyme disease is often difficult to diagnose because its symptoms and signs mimic those of many other diseases, such as influenza, infectious mononucleosis, rheumatoid arthritis, multiple sclerosis<sup>3,5</sup>. Laboratory diagnostics is suboptimal, meaning that that the laboratory tests are positive only in 40–60% of the cases. Serology in use is as follows: Imunoflorescens Assay or Antibody titer (IFA), Enzyme Linked Immunosorbent Assay (ELISA), Immunoblot or Western blot (WB) for the IgG and IgM antibodies. As serodiagnostic screening method for Lyme borreliosis, ELISA alone or in combination with Western blot method is used.

Above mentioned tests are more useful in later stage of the disease, but even then the results can be incorrect. In early stage I, Lyme IgM ELISA will be negative<sup>12</sup>.

Regarding therapy, following antibiotics are effective: tetracycline, erythromycin, penicillin, and cephalosporin. Patients treated in the early stages with antibiotics usually recover rapidly and completely. Varying degrees of permanent damage to joints or the nervous system can develop in patients with late chronic Lyme disease<sup>3,5,6</sup>. Typically these are patients in whom Lyme disease was unrecognized in the early stages or for whom the initial treatment was unsuccessful. Rare deaths from Lyme disease have been reported.

## Patients and Methods

### Case report

A 48-years old female patient hospitalized in our clinic due to a surgery of paralytic strabismus as a late consequence of the Lyme disease. In September of 1985 the patient was hospitalized to Neurology clinic because of headache, dizziness and diplopia. The symptoms began in March of the same year when the patient suffered some kind of viral disease. For a five months period the patient had weakness, dizziness, and headache. A thorough examination discovered vertebral column changes. Rehabilitation was suggested, but without any improvement. The patient experienced even stronger dizziness, troublesome walking with gait moving towards left side. Pain in the neck spread towards shoulder region; diplopia was still present as were hearing troubles. Complete neurological examination was performed including brain CT scan that showed no pathol-

ogy. For further test, the patient was admitted to hospital. Romberg was positive, especially to the left side; there was horizontal nystagmus, right side gaze diplopia and hyperesthesia of the left side of the face. Control CT scan was within normal parameters. An infectious disease specialist was consulted; lumbar puncture was performed but no signs of toxoplasmosis, listeria or tics transmitted encephalitis were found. The patient was released from hospital with the diagnose of post viral encephalitis and suspicion of multiple sclerosis.

The same year, in October, left knee and swelling were added to the persisting clinical symptoms. Because of that, in February of 1986, the patient underwent examination at the Infectious disease Clinic. Horizontal nystagmus with diplopia, hypalgesia of the right side of the face with periphery type of facial nerve paresis were found. Lyme disease was suspected and laboratory test were performed as well as serology on *Borrelia burgdorferi*. The results showed positive serum IgM titer and negative IgG. CSF analysis showed IgM negative and IgG positive results. Patient's history revealed tic bite of the left arm in March of 1985 with no skin changes. Hospitalization was suggested with immediate 20 million i.u. i.v. Penicillin treatment during 12 days with 2.4 million i.u. i.m. Extencillin on the 13<sup>th</sup> day. Some improvement was noticed: neurological symptoms were reducing, as well as dizziness and headache but neck pain and thoracic vertebrae pain persisted.

Fundoscopy examination of both eyes showed discreetly pale optic nerve head of the temporal side with bilateral visual acuity 1.0. Prescribed eyeglasses with prisms were not satisfactory. In the next two years the patient was hospitalized two more times at the Infectious disease clinic. In February of 1988 psychology results indicated organic mental disorder. In March of 1988 a series of test were performed in Audiological center due to instability. A spontaneous and left gaze oriented nystagmus was found as well as vestibulospinal abnormalities. ENG was performed. Vertigo centrale and Laesio vestibularis centralis were diagnosed. In March of 1992 during invalid retirement procedure, a routine CT scan showed cerebellum atrophy with hypodensity in pons and medial parts of temporal lobe (vertebro-basilar insufficiency). Disseminated recidivated encephalomyelitis signs were discovered and MRI scan was advised that confirmed the diagnosis. In September of 1993 hospitalization to Eye clinic due to significant bilateral loss of vision. Visual field testing showed concentric narrowing of inner isopters. Hess Lancaster test: paresis nervi abducentis lateris dextri et musculus obliquus inferioris oculi dextri. With corticosteroid therapy in an infusion and vitamins per os, bilateral visual acuity improved to 1.0. Surgical treatment of paralytic strabismus was not advised, but in therapy vitamins were recommended and paralytic eye occlusion to avoid diplopia. New prism eyeglasses were prescribed. In April of 1996 the patient was examined in our clinic, and paralytic strabismus surgery was recommended.

Two years later the advised surgical procedure was performed. At the time of operation, the squint angle was +40, paralysis of abductal nerve of the right eye without any temporal and with limited up and down movement. Fundoscopic examination showed optical nerve head to be pale temporal. The operating procedure was performed under general anesthesia with reposition of right internal straight muscle and resection of external straight muscle of the same eye.

The operating procedure was well tolerated. Squint angle was reduced to +10, but still without any temporal movement. Diplopia was still present but reasonable decreased with the use prism eyeglasses. The patient was satisfied with the cosmetic outcome and quality of life, generally improved.

## Discussion

Lyme disease is one of the most common zoonosis in Europe and United States. It is a multi system disorder caused by *Borrelia burgdorferi* spirochete<sup>2-4</sup>. The diagnosis is based on a several clinical criteria spread over three stages of the disease and supportive data from laboratory testing<sup>12-14</sup>. It is known to have a variety of

neurological symptoms which include ocular manifestations all three stages of disease<sup>1,15,16</sup>. Positive endemic area history is the anamnestic data of most importance<sup>1</sup>. Such patients should be serologically tested on *Borrelia burgdorferi* in order to confirm the exact diagnosis<sup>12,16</sup>. With clinical symptoms and serological results, oral antibiotics therapy should be used<sup>16</sup>. When anterior eye segment is involved, topical corticosteroids should be used. Intravenous antibiotics should be used in posterior eye segment involvement, and together with corticosteroids in neuroophthalmic disease. When ocular manifestations are detected in early stage, almost complete restitution is possible. That makes serology tests necessary in all patients with unknown neuroophthalmic diagnose<sup>6,12,16</sup>. In our case, since the real diagnosis was established in the late stage of the disease, only partial recovery was achieved. In summary, although orbital involvement of *Borrelia burgdorferi* is an unlikely occurrence, with appropriate clinical presentation, and lacking evidence of other diseases, Lyme disease should be taken into consideration. With positive serology tests and imaging techniques, the diagnose could be established.

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## PARALITIČKI STRABIZAM KAO MANIFESTACIJA LYME BORRELIOZE

### SAŽETAK

Lyme borelioza je multisistemska bolest uzrokovana spirohetom *Borrelia burgdorferi*. Očne manifestacije su opisane, no njihova pojavnost je vrlo rijetka. Predmet našeg rada jest prikaz pacijentice stare 49 godina koju je 1985. godine ugrizao krpelj te je kao posljedicu istog razvila simptome Lyme borelioze. Pacijentica je 1998. godine hospitalizirana na našoj Klinici radi operativnog liječenja paralitičkog strabizma (paraliza n.abducensa), rijetke očne manifestacije Lyme borelioze. Postoperativno je kut škiljenja znatno reduciran, no još uvijek bez pokretljivosti bulbosa temporalno. Dvoslike su još uvijek prisutne, no značajno smanjene korekcijom s prizmama. Postignuto je zadovoljstvo pacijenta uz poboljšanje kvalitete života.