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Case Report

Lyme Neuroborreliosis Presenting with Symptoms of Night Terrors: A Case Report

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Abstract

This case describes an unusual presentation of Lyme neuroborreliosis in a child presenting with nightly episodes of screaming and crying followed by amnesia for the event.

ABBREVIATIONS

CSF: Cerebrospinal F; CXCL13: Chemokine CXC motif ligand 13; EEG: Electroencephalogram; LNB: Lyme Neuroborreliosis; PCR: Polymerase Chain Reaction

INTRODUCTION

Lyme borreliosis is the most common vector-borne disease in Europe [1,2], with a large number of cases occurring in children [2]. It is caused by *Borrelia burgdorferi sensu lato complex* spirochetes, which is transmitted to humans through bites from infected ticks [3]. Lyme borreliosis is a multisystem syndrome affecting the skin, joints, heart, and nervous system in different stages of the disease [3].

Lyme neuroborreliosis (LNB) is among the most prevalent bacterial infections of the nervous system. In Denmark an annual incidence rate of 0.7-4.7 per 100,000 inhabitants has been reported with children comprising approximately 20% of the cases. In children, LNB typically presents with acute facial nerve palsy and/or subacute meningitis [4-6]. Unspecific but common symptoms are headache, loss of appetite, fatigue, nausea, and unspecific pain [7].

This case describes LNB presenting with symptoms of night terrors in a child.

CASE PRESENTATION

A 5-year-old, previously healthy, girl presented in August 2020 with a 14-day history of nightly screaming, crying and complaints about headache and neck pain. Symptoms occurred 2 to 3 times per hour and lasted a few minutes followed by spontaneous cessation. In the morning the girl was in her usual happy mood and did not remember what had happened during the night. At daytime she felt well except for being tired due to interrupted sleep. Furthermore, upon asking, the last two month she had had episodes of neck pain, arthralgias and increased sensitivity to touch and had consulted a chiropractor. However, she had continued her everyday life. Additionally, the family recalled days with sub febrile temperature without known cause. She was attending a kindergarten in the forest north of Copenhagen and her parents recalled that she had had a tick bite 2 month prior to admission; the tick was removed < 24 hours after the bite. No erythema migrans was observed.

At initial admission, the girl's physical and neurological examination was normal, namely with no fever or neck stiffness. Basic lab work, including white blood count was within normal range. Due to acute onset of an unusual presentation of night terror a Computed Tomography (CT) scan of the head was performed, normal. EEG including sleep was normal with no paroxysmal activity. One week later her symptoms had not ceased, so a lumbar puncture was performed due to suspicion of LNB. The cerebrospinal fluid (CSF) showed CSF-pleocytosis with white blood cells: 177 E6/L, (normal range < 5 E6/L) of which 67% were mononuclear cells, elevated CSF-protein: 1.92 g/L (normal range: 0.15-0.45 g/L) and normal glucose 2.5 mmol/L (normal range: 2.2-3.9 mmol/L),

Point of Care (POC) test of CSF chemokine CXC motif ligand 13 (CXCL13) was markedly elevated 430 ng/L (< 250 ng/L).

The CSF was cultured, negative. Following agents were excluded by a quick polymerase chain reaction (PCR) test: *Enterovirus, parechovirus, human herpesvirus 6A/B cytomegalovirus, herpes simplex virus and varicella zoster virus*

As the girl had CSF pleocytosis and the CXCL13 was markedly elevated LNB was suspected and the girl was started on orally doxycycline 200 mg BID for 14 days. The patient's symptoms were reduced already the first night after antibiotic therapy was

initiated and she had completely recovered after 48 hours of therapy.

Intrathecal *B.burgdorferi* antibody index IgG/IgM was positive 27.5/3.8) (Normal value <= 0.3).

DISCUSSION

LNB is the most common manifestation of dissemination of Lyme borreliosis in Europe [8]. It presents with a wide spectrum of clinical manifestations, of which aseptic meningitis and cranial nerve palsies are the most common manifestations in children [4,5]. Night terror, however, is a rare clinical presentation which to our knowledge has only been mentioned in two previous studies [9,10].

A night terror is characterized by autonomic nervous system activation, including tachycardia and tachypnea, along with facial expressions of intense fear, screaming, shouting and agitation [11]. It is considered a parasomnia associated with non-rapid eye movement (non-REM) sleep stages in which the child is in a transitional state between sleep and wakefulness. In the morning the child has amnesia from the event. The exact etiology of night terrors is unknown. Developmental, environmental, psychological, and genetic factors have been identified as potential causes [12]. Furthermore, a correlation between night terrors and fever/illness has been suggested [13,14]. It is uncertain if our case presented with night terrors or with symptoms mimicking night terrors.

Although most children with LNB present with cranial nerve palsies, atypical presentations without neurological signs may occur [6]. A register-based study on 89 children with LNB found that 79% had one or more objective neurological signs at presentation, most frequently facial palsy. Twenty-one percent, however, had no neurological signs at physical examination. In these children diagnostic delay was longer compared with those with objective neurological abnormalities [6].

Due to the diverse clinical manifestations of LNB, laboratory investigations are used to confirm the diagnosis. Pleocytosis in CSF as well as serum and CSF antibodies against *B.burgdorferi* are most commonly present in patients with LNB [15]. However, the intrathecal antibody production does not start immediately after dissemination to the nervous system. Thus anti-*B. burgdorferi* antibodies may still be absent if lumbar puncture is performed early in the course of the disease [15]. The chemokine CXCL13 has been identified as an early marker for LNB in children [15]. CXCL13 is produced upon detection of intrathecal spirochetes by monocytes, dendritic cells and numerous other cell types and is a key factor for B-cell immigration into the CSF in LNB. Thus, the production of CXCL13 precedes the antibody synthesis [15]. A prospective case-control study found that CXCL13 was elevated in 73% of children with LNB and pleocytosis, but without detectable intrathecally produced anti-*B.burgdorferi* antibodies. Additionally, CXCL13 showed both high sensitivity (88%) and specificity (89%) in LNB in children [15]. Besides facilitating early detection, CXCL13 may be a helpful marker to strengthen the diagnosis of LNB in children with unusual presentations, as in the case presented here. Further, the result of a CXCL13 POC can be ready within one hour whereas Borrelia serology testing may not be performed on daily basis as in our laboratory.

Although rare, LNB should be considered in children presenting with symptoms of night terrors, particularly in Lyme-endemic areas. Along with the presence of symptoms, the diagnosis of LNB can be supported by lumbar puncture findings including mononuclear CSF pleocytosis. Furthermore, the use of the chemokine CXCL13 can be beneficial in early detection of LNB in children.

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