

CLINICAL NOTE

Lyme Disease Presenting as Isolated Acute Urinary Retention Caused by Transverse Myelitis: An Electrophysiological and Urodynamical Study

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ABSTRACT. Olivares JP, Pellas F, Ceccaldi M, Viton JM, Raoult D, Planche D, Delarque A. Lyme disease presenting as isolated acute urinary retention caused by transverse myelitis: an electrophysiological and urodynamical study. *Arch Phys Med Rehabil* 1995;76:1171-2.

Several neurological manifestations of Lyme disease, both central and peripheral, have been described.¹ Reported here is a case of acute transverse myelitis related to a Lyme neuroborreliosis that presented with isolated acute urinary retention and no lower-extremity impairment. This case, documented by urodynamic and electrophysiological investigations, partially resolved after 6 weeks of intravenous ceftriaxone, affording the removal of the indwelling catheter. Alpha blocker therapy was needed for 3 months, until the complete normalisation of urodynamic and electrophysiological records. This case study indicates that whenever urinary retention is encountered associated with acute transverse myelitis or alone, the patient should be investigated for Lyme disease.

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ACUTE TRANSVERSE MYELITIS (ATM) has been reported after infections,¹ as well as vascular, auto-immune, traumatic, and space-occupying disorders. The postinfectious manifestation of ATM is a classical etiology of endemic occurrence in the population. In contrast, acute transverse myelopathy is a rare complication of viral (human immunodeficiency virus [HIV], cytomegalovirus, herpes simplex virus, rubella, chickenpox, infectious mononucleosis, measles)² and spirochetal infections^{3,4} as well as tuberculosis.² ATM after Lyme borreliosis is rare and infrequently reported in the medical literature,^{1,3} although bladder dysfunction is common in Lyme disease. Reported here is a case of Lyme neuroborreliosis that presented with isolated acute urinary retention.

CASE REPORT

A 54-year-old man was initially referred for acute urinary retention by his general practitioner to the urology department; he had suffered from dysuria and burning micturition for 2 months. On clinical admission, body temperature was normal, but bilateral hydronephrosis with no apparent urological cause was discovered.

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Referred to the neurology department, acute myelitis was immediately discussed and investigated: neurological examination of the lower limbs showed bilateral extensor plantar response and deep tendon (knee jerk - L4 - and ankle jerk - S1 -) hyperreflexia, without muscle hypertonia or weakness, nor any sensory impairment: recognition of toe and foot movements, as well as perception of vibrations, were correct. As a result, the gait walking pattern was normal. The rest of the neurological examination of the upper limbs was normal and no cranial nerve or cerebellar deficits were observed. No other systemic manifestations of Lyme disease were evident during the examination or as part of the patient medical history.

Erythrocyte sedimentation rate was 95, white blood cell count showed 11,400/mL white cells (63% neutrophils), and serology for hepatitis B, HIV virus, syphilis, brucellosis, rickettsiosis, and hydatidosis was all negative. Examination of sputum and the gastric washings were negative for tuberculosis. Lumbar puncture performed 2 weeks after admission showed a clear cerebrospinal fluid (CSF), containing two red blood cells without leukocytosis and a moderately elevated level of protein (79mg/dL) and immunoglobulin G (10.6mg/dL).

Visual and brainstem auditory evoked potentials (EV) were normal, whereas the magnitude of the somesthetic EV at the lower limbs was reduced. Computerized tomography, magnetic resonance imaging (MRI) of the spinal cord, and myelography were normal.

Lyme titers performed by the indirect immunofluorescence technique were positive for both blood (Ig G threshold at 1 to 100 and Ig M threshold at 1 to 50) and cerebrospinal fluid (no threshold, any positive titer indicates the absolute presence of the disease). One week after admission, the Ig G blood titer was at 1 to 400 and IgM at 1 to 100, whereas 2 weeks after admission the Ig G blood titer was at 1 to 800 and IgM at 1 to 25. These findings were confirmed by western blot technique.

After this examination, the patient was further referred to the department of rehabilitation to investigate associated urinary signs: marked striated sphincter anal hypertonia and a decreased pinprick sensation of S3-S4 perianal region. Urinary retention was initially managed with an indwelling Foley catheter.

Urodynamic evaluation 1 week after hospitalization showed, before treatment, reduced bladder compliance, normal bladder sensitivity, increased striated sphincter closing pressure at 170cm/H₂O (normal range = (110 - age) ± 20% = 56 ± 11cm/H₂O), and a detrusor/external sphincter dyssynergia. Somatosensory evoked potentials (after pudendal nerve stimulation) demonstrated an increased P40 (50.4ms) and N50 (58.6ms), whereas electromyography (EMG) of the striated sphincter and of the bulbo-cavernous muscles showed pseudomyotonic discharges and a fast firing rate. The latency of the left balano-urethral reflex was slightly elevated at 45.4ms (normal below 35.5 ± 9ms), whereas the left bulbo-cavernous reflex had a normal latency.

After the diagnosis of Lyme disease, the neurology department prescribed conventional antibiotic treatment for a 6-weeks period, first by intravenous Ceftriaxone (1g twice a day for 4

weeks) then with oral Doxycycline (200mg/day) for the next 2 weeks. At the end of the 6-week antibiotic treatment, checking of the urodynamic and the EMG examinations showed a less increased striated sphincter closing pressure (110cm/H₂O) with a still reduced bladder compliance, no denervation on the electromyogram of the striated sphincter, and no detrusor/external sphincter dyssynergia. The pudendal evoked potential, as well as the left balano-urethral and bulbocavernous reflexes were normal. Control of serum Lyme titers showed a reduced positivity for IgG (at 1 to 200) and IgM (at 0). The indwelling Foley catheter was permanently removed, and the patient was then taught intermittent self-catheterization. The neurological examination was normal. At the completion of their investigations, the rehabilitation department in conjunction with the neurology department prescribed alpha blocker therapy with Moxisylyte (carlitene),^a 270mg/day, for the next 3 months, after which time the patient was able to urinate normally but refused any further urodynamic or EMG investigation.

DISCUSSION

The spirochete *Borrelia burgdorferi* that had been identified as the cause of Lyme disease is predominately transmitted by the tick.⁵ Steere⁴ suggested a temporal classification of clinical findings of Lyme disease into three chronological stages starting from the original tick bite. *Stage 1* includes the initial characteristic cutaneous rash (erythema chronicum migrans); other systemic symptoms or positive blood tests may be present but frequently are not noticed until the next stage. *Stage 2*, months later, is characterized by evident systemic symptoms with neurological and cardiac manifestations, as well as high titers of Lyme antibody. At this time, the "classic triad" of the so-called "neuroborreliosis" (aseptic meningitis, peripheral radiculoneuritis, and cranial palsies) occur in 10% to 20% of the untreated patients.³ *Stage 3*, years later, has initially been classified by the classical occurrence of arthritis.

It now becomes evident that during stage 3 central neurological manifestations may be observed, even if they are not the most usual signs. Meningitis, cranial neuritis, and radiculoneuritis are the most frequently reported nervous system manifestations of Lyme disease at this stage.^{1,3,6}

Considering both peripheral and central manifestations, neuroborreliosis, can occur both during the second and third stages of the disease. Peripheral nervous system manifestations of neuroborreliosis (classic triad) are its unique expression during stage 2, whereas transverse myelitis^{3,7,8} and other central manifestations⁹ like encephalopathy or psychiatric features can be associated with stage 3.

Thus, in Lyme Borreliosis, transverse myelitis is noted only in some reported cases.^{3,7,8} In those cases, it becomes the disease's prominent expression, demonstrated by spasmodic tetraparesis,⁷ paraparesis alone,⁸ or associated urinary retention.³

In another case of neuroborreliosis reported with bladder dysfunction,³ during the first days after onset of the disease as well as at its end when bladder dysfunction resolved after paralysis, no other neurological abnormality was present. The second relevant observation with this patient was the occurrence, 5 years previously, of a Bell's palsy resolving spontaneously. Such a long delay between the first peripheral neurological manifestation and the actual central symptoms of ATM led the authors to consider their case to be in its *third stage*. At that time, after 6 weeks of antibiotics, improvement progressed from paralysis to walking, but the patient still had a sensory deficit consisting of a tingling and numbness sensation in the legs.

In this case, very similar temporal occurrences were described for the perineal manifestations and lower limbs neurological

symptoms, with urological dysfunction preceding them and later resolving. Some discrepancies therefore occur between this case and the reported case.

First of all, considering the extremely moderate intensity of lower limb neurological manifestations—Babinski's sign and brisk reflexes—it is assumed that this case is the first one in which urinary retention was the unique evident and permanent clinical expression of an acute transverse myelitis caused by Lyme Borreliosis, and to the authors' knowledge represents the first one documented by electromyogram of the striated sphincter, evoked potentials and urodynamics.

The second discrepancy concerns the difference in temporal presentations of the disease and its responsiveness to the treatment between the two cases. The other reported case was probably an old stage 3 Lyme disease, considering the history of cranial palsy 5 years earlier, the high intensity of central manifestations, and the remaining posterior column dysfunctions after prolonged antibiotics. On the other hand, this patient, presented on admission with only recent urological symptoms of ATM. His medical history was free of any of the classic components of the "neuroborreliosis triad" (aseptic meningitis, peripheral radiculoneuritis, or cranial palsy). He presented high titers of Lyme antibody, and all ATM manifestations completely resolved after antibiotic treatment.

This case is an unusual one, in as much as considering the first two arguments it should be considered as a stage 3 of the disease, in which all the second stage had been silent. However, considering the high titers of Lyme antibody and the good response to antibiotics, it should be classified as a stage 2. On the contrary, assuming that ATM could be observed early at stage 2—in the absence of cardiac symptoms or of neuroborreliosis triad—it could be suggested classifying this patient as a stage 2 case, whose central system manifestations resolved completely after antibiotic treatment.

This case study should prompt all physicians who encounter urinary retention in their practice to scrupulously investigate for Lyme disease, even if no sensory or motor impairment of limbs are noticed, and if there are no elements of the classic neuroborreliosis triad in the medical history. Because Lyme borreliosis is a health problem of increasing international importance and can be potentially reversible on appropriate antibiotic therapy, any physician must be aware that an isolated urinary dysfunction can be the unique manifestation of Lyme Borreliosis even outside an endemic area.

References

1. Finkel MF. Lyme disease and its neurological complications. *Arch Neurol* 1988;45:99-104.
2. Linssen WHJ, Gabreels FJM, Wevers RA. Infective acute transverse myelopathy. A report of two cases. *Neuropediatrics* 1991;22:107-9.
3. Chancellor MB, Dato VM, Yang GY. Lyme disease presenting as urinary retention. *J Urol* 1990;143:1223-4.
4. Steere AC, Grodzicki RL, Kornblatt AN, Craft JE, Barbour AG, Burgdorfer W. The spirochetal etiology of Lyme disease. *N Engl J Med* 1983;308:733-40.
5. Burgdorfer W, Barbour AG, Hayes SF, et al. Lyme disease: a tick-borne spirochetosis? *Science* 1982;216:1317.
6. Reik L, Steere AC, Bartenhagen NH, et al. Neurologic abnormalities of Lyme disease. *Medicine* 1979;58:281-94.
7. Kohler J. Lyme borreliosis: a case of transverse myelitis with syrinx cavity. *Neurology* 1989;39:1553-4.
8. Rousseau JJ, Lust C, Zangerle P, et al. Acute transverse myelitis as presenting neurological feature of Lyme disease. *Lancet* 1986; 2:1222-3.
9. Steere AC. Lyme disease. *N Engl J Med* 1989;321:586.

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