This is a report of 2 patients with Lyme disease who initially presented with severe constipation, which progressed to ascending muscular weakness resembling acute idiopathic polyneuritis, with neuropsychiatric symptoms, severe urinary retention, and hyponatremia. These symptoms resolved following proper antibiotic therapy.

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Lyme disease is the most common preventable vector-borne illness in the United States, and *Borrelia burgdorferi* is the causative agent. This infectious agent is transmitted to humans by the bite of the *Ixodes* tick, and was discovered in a group of children with arthritis in Lyme, Conn, in 1975. In the United States, the disease is polarized largely to the northeastern regions, and nearly 15,000 cases are reported annually to the Centers for Disease Control and Prevention.

The clinical manifestations of acute Lyme disease are multisystemic and involve the nervous system. Approximately 17% of those affected have neurologic manifestations as early signs, with meningeal irritation (17%), headache (64%), neck stiffness (48%), arthralgias (48%), myalgias (43%), backache (26%), dysesthesias (11%), photophobia (6%), and dizziness (5%). Unilateral and bilateral facial paralysis may occur in up to 11% of patients with Lyme disease.

Herein, we describe 2 patients with a similar and atypical clinical presentation of Lyme disease with severe constipation, followed by a syndrome similar to acute idiopathic polyneuritis, sensory deficits, urinary retention, hyponatremia, and visual hallucinations.

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tact, and his pupils were round and reactive to light; he had a corrected visual acuity of 20/30 OU. No visual field or color vision deficits were noted. Cranial nerves IX through XII were intact. He had mild weakness in the proximal upper extremities and profound distal weakness that hampered his grasping a glass with his hands and writing. In the lower extremities, his strength was especially decreased proximally. He was unable to bear weight on his legs. Atrophy was not present. There was sensory loss in both hands that extended to the right forearm but spared the left. There was reduced sensation bilaterally on his torso at the level of the sixth and seventh thoracic vertebrae. All tendon reflexes were absent, with equivocal plantar responses bilaterally.

He developed visual hallucinations consisting of snakes climbing up the wall that he described with his eyes open. They would disappear on the ceiling, and new ones would appear on the wall. These images did not scare the patient, but were intriguing to him. The presence of people in the room did not interfere with the images, which disappeared spontaneously after 2 days.

Laboratory findings included an initial white blood cell count of 11.1 × 10³/µL, which increased to 18.4 × 10³/µL, with 82% granulocytes. His platelet count was initially at 388 × 10³/µL, and ranged as high as 424 × 10³/µL. His serum sodium level was 125 mEq/L. A cerebrospinal fluid (CSF) analysis revealed a red blood cell count of 24/µL and a white blood cell count of 480/µL, with 100% lymphocytes; a total protein level of 0.47 g/dL; and a glucose level of 39 mg/dL (2.16 mmol/L). The lymphocytes were atypical; cytologic and flow cytometric evaluations described them as inflammatory. In the serum and CSF, IgM titers were positive for Lyme disease, and a Western blot analysis of the CSF confirmed Lyme disease. The electromyographic results were consistent with acute demyelinating polyradiculopathy. The result of a cardiac examination was unremarkable.

Within days of starting intravenous ceftriaxone therapy, the patient’s serum and CSF laboratory values normalized, the hallucinations disappeared, and the constipation resolved; he also regained strength in his lower extremities. Hyponatremia also resolved after fluid restriction. With physical therapy, he fully recovered in 6 months.

CASE 2

An 84-year-old white man who presented with severe constipation, refractory to medical treatment, followed 2 weeks later by weakness and numbness that started in the right anterior thigh and upper shin and progressed to the left lower extremity. Apart from generalized fatigue and dyspnea, he complained of urinary retention.

Two weeks before the onset of the constipation, a rash on his hand was treated with cephalaxin, with no history of an insect bite. He was initially admitted to the intensive care unit and treated with intravenous immunoglobulin, with the presumptive diagnosis of acute idiopathic polyneuritis, without resolution of these symptoms. On hospital admission, his vital signs were normal. The results of the cardiopulmonary and abdominal examinations were unremarkable, and his bowel sounds were hypoactive. He was alert and oriented, with a decreased attention span. His speech was fluent, with good comprehension, intact naming, and repetition. He had no strength in his lower extremities, but could wiggle his toes. There was only mild weakness in the upper extremities and no atrophy. Reflexes were normal in the upper extremities but absent in the lower extremities, with flexor plantar responses. Sensation was diminished to all modalities in the lower extremities, and intact in the upper extremities. Cranial nerves II through XII were intact, and his right eye was prothetic.

His white blood cell count was 14.5 × 10³/µL, with granulocyte predominance; his platelet count was 306 × 10³/µL. He was also hyponatremic (sodium level, 121 mEq/L). His CSF protein level was 0.18 g/dL, his glucose level was 44 mg/dL (2.44 mmol/L), and his CSF white blood cell count was 282/µL, with lymphocytic predominance (94%) and no red blood cells. Cerebrospinal fluid and serum titers were positive for Lyme disease, with confirmation by CSF Western blot analysis. The electromyographic results were consistent with polyradiculopathy. The result of a cardiac examination was unremarkable.

He had hallucinations of unknown people walking in and out of the room, and was able to see them with his right prosthetic eye, which he knew was impossible. He also felt depressed and wanted to die. Urinary retention was severe enough to require a Foley catheter, and constipation continued.

Within days of starting intravenous ceftriaxone therapy, the patient’s serum and CSF laboratory values normalized, the hallucinations disappeared, and the constipation resolved; he also regained strength in his lower extremities. Hyponatremia also resolved after fluid restriction. With physical therapy, he fully recovered in 6 months.

COMMENT

Our 2 patients had documented Lyme disease manifesting initially with autonomic symptoms: severe constipation and urinary retention progressing to a syndrome similar to acute idiopathic polyneuritis, with hallucinations and hyponatremia. Both had a history of an atypical rash without central clearing. The presence of an atypical rash in patients with Lyme disease, however, has been described in as many as 50% of the cases.6

In the classic description of the clinical manifestations of Lyme disease, gastrointestinal tract symptoms, excluding constipation, were observed in 10% of patients.3 This is also the case with other spirochetal infections, in which constipation is not a characteristic symptom.7 Within days after starting antibiotic treatment, both patients’ constipation resolved. The pathophysiological features of the constipation in our patients are unclear, but rapid resolution with antibiotics strongly suggests that this symptom was directly produced by the infection. Neuropsychiatric manifestations of acute neuroborreliosis, including hallucinations, psychoses, and encephalopathy, have been documented,4,4 but their cause is unclear. In case 1, there was no obvious underlying psychiatric history and other organic causes could not explain the hallucinations. They resolved with the start
of antibiotic therapy and were thought to be at least temporally associated with the borreliosis. We are unable to explain the cause of the hallucinations, but speculate that neuroborreliosis could be the cause. Lyme psychosis has been reported mainly in Europe, where the genotype, *B burgdorferi* sensu stricto, is associated with Lyme disease. It is likely that this genotypic difference leads to varying phenotypic expressions of the disease.

It is hard to attribute the hallucinations simply to the hyponatremia, because only 0.5% of patients with hyponatremia exhibit hallucinations and usually at serum sodium levels below 120 mEq/L (an average of 115 mEq/L). Psychiatric manifestations of Lyme disease have been reported for many years. Although exact numbers are not available, depression is reported in 26% to 66% of patients with Lyme disease. Multiple neuropsychiatric symptoms have been linked to Lyme disease, including visual hallucinations.

Urinary retention was once described as an initial presentation of Lyme disease. In that patient, serum Lyme titers were high, which likely resulted from tertiary Lyme disease with a history of untreated Bell palsy 5 years earlier. In our patients, the symptom was associated with acute neuroborreliosis, and improved following antibiotic treatment. The constipation and urinary retention may reflect involvement of the autonomic peripheral nervous system.

The hyponatremia exhibited by both patients was considered, following a complete endocrine examination, to be the result of the syndrome of inappropriate antidiuretic hormone. To our knowledge, this has not been reported previously in a patient with Lyme disease, but inflammatory and infectious central nervous system processes have been associated with the syndrome of inappropriate antidiuretic hormone, including acute idiopathic polynuromyelitis and meningitis. In the latter, fungal, bacterial, carcinomatous, and tuberculous infections have been implicated. In our patients, fluid restriction was instituted, with a good response.

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REFERENCES