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Case Report

Abdominal Wall Weakness and Lumboabdominal Pain Revealing Neuroborreliosis: A Report of Three Cases

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Abstract: The authors report three cases of thoracic radiculoneuropathy disclosing neuroborreliosis. All three patients had low back and abdominal pain and two had marked abdominal wall paresis. EMG confirmed a motor involvement of the lower thoracic roots and CSF analysis revealed a lymphocytic meningitis in all three cases. Antibodies against *Borrelia burgdorferi* were present in both the serum and the CSF. A favourable outcome was obtained in all three patients with appropriate antibiotherapy. The differential diagnosis of this misleading presentation is discussed.

Keywords: Abdominal muscle paralysis; Abdominal pain; Low back pain; Lyme neuroborreliosis; Thoracic polyradiculoneuropathy

Introduction

Stage II neuroborreliosis is common in Lyme disease, occurring in up to 40% of patients. The most frequent features are lymphocytic meningitis, radiculoneuritis and cranial neuropathy (often seventh nerve palsy), either alone or in combination: the so-called Bannwarth syndrome [1]. Radiculoneuropathy usually starts as a painful limb with dysaesthesia and a variable degree of paresis. Initial isolated thoracic radiculoneuropathy is rare. We report three cases of Lyme disease with the unusual presenting features of abdominal and low back pain, in addition to a striking abdominal distension in two patients.

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Case Reports

Case 1

A 54-year-old man with no significant medical history developed continuous low back pain radiating along the waist on both sides, unrelated to motion or previous effort. The pain was maximal at the end of the night, forcing him to get up and walk about. One week later, he noted rapidly progressive abdominal wall distension and complained of constipation. Some usual gymnastic exercises became impossible owing to abdominal muscle paresis. Bowel X-rays and abdominal echography were normal. One month later he was admitted to the rheumatology department when a left facial palsy appeared. He could not recall any tick bite or erythematous rash. Physical examination revealed a protuberant abdomen with diffuse superficial pain, but without organomegaly. Knee jerks were absent, but all other deep tendon reflexes were brisk. Superficial abdominal reflexes could not be elicited. Sensation was impaired for light touch and pinprick in the T10–L1 dermatomes and in the left L4 dermatome. There was a left seventh nerve palsy. ELISA was positive for serum IgG antibodies against *Borrelia burgdorferi*. CSF contained 130 white blood cells/mm^3 (86% lymphocytes). The CSF protein was 188 mg/dl and glucose 40 mg/dl. IgG antibodies against *Borrelia burgdorferi* were also detected in the CSF. Immunoblotting revealed eight specific IgG oligoclonal bands with antibody activity against *Borrelia burgdorferi*. Other laboratory tests were normal or negative, including varicella zoster serology. Electromyography showed active denervation (fibrillation potentials and positive sharp waves) in the rectus abdominis muscles at the T11 level and in the muscles innervated by the right L4 and left L4 and L5 roots. The

patient was treated with intravenous penicillin, 20.10^6 U a day for 1 month. Four months later, sensation in the left L4 root territory was still slightly impaired and knee jerks were weak. The pain had disappeared, as well as the abdominal wall paresis.

Case 2

This 56-year-old man, with a past medical history of hypertension, type II diabetes, hypercholesterolemia, urinary lithiasis and gastritis, developed acute and severe nocturnal low back pain, radiating along both sides of the abdomen to the groins and umbilicus. The pain was not increased by movement or effort. At the same time, he noticed a sensation of abdominal distension. After the second day he was admitted to the emergency room, where urinary lithiasis was ruled out and a CT scan of the abdomen was normal. One month earlier he had suffered from a right sciatalgia that resolved after a 10-day course of non-steroidal anti-inflammatories. He reported a possible tick bite 3 months earlier, but without an erythematous rash. He was admitted to the rheumatology department. Clinical examination revealed a moderate abdominal distension and hypoaesthesia for light touch and pinprick over the abdomen from the T10 to T12 on both sides. Sensation was normal over the back. Superficial abdominal reflexes were absent. Mobility of the spine and paravertebral muscle tone were normal. A marked paresis of abdominal muscles with an important distension of the abdominal wall appeared progressively over the subsequent days, with no modification of body weight or bowel habits. Serum antibodies against *Borrelia burgdorferi* tested by ELISA were positive for IgG and negative for IgM. Diabetes was well controlled. CSF analysis revealed 138 white blood cells/mm^3 (80% lymphocytes, 15% monocytes), total protein of 119 mg/dl, glucose of 93 mg/dl and positive IgG antibodies against *Borrelia burgdorferi*. Immunoblotting revealed specific IgG oligoclonal bands. EMG showed active denervation in the abdominal and paraspinal muscles at T10 and T11 levels on both sides. X-rays and MRI of the spine were normal except for a mild L4/L5 discopathy. He was treated with ceftriaxone, 2 g/day for 14 days. Four months later the neurological examination was normal.

Case 3

This 61-year-old woman had a medical history of hypothyroidism, hypertension, smoking, asthma and lumboabdominal zoster which had occurred 1 year before admission and was completely resolved. She was admitted to the gastroenterology department after several days of continuous diffuse abdominal pain, with superficial burning sensations associated with continuous low back pain. There was no skin lesion at that time but she could remember being bitten in the right thigh 1 month earlier. This was rapidly followed by an erythematous and painless rash in the same area, with progressive extension and complete resolution within 2 weeks. Clinical examination disclosed a suspended bilateral sensory loss to pinprick and light touch, with dysesthesia from T10 to L1. Superficial abdominal reflexes could not be elicited, but the patient was very obese. For the same reason, it was impossible to ascertain abdominal muscle strength. Otherwise, examination of the abdomen was normal and it did not seem unusually protuberant to her nor to her family. The right ankle jerk was absent. Other deep tendon reflexes were weak. Mobility of the spine was normal. Serum IgM antibodies against *Borrelia burgdorferi* were present but no IgG were detected by ELISA. CSF contained 600 white blood cell/mm^3 (80% lymphocytes, 10% monocytes). The CSF protein was 242 mg/dl and glucose 59 mg/dl. Both IgG and IgM antibodies against *Borrelia burgdorferi* were found in the CSF, and immunoblotting revealed several specific IgG oligoclonal bands. Abdominal echography and CT scan were normal. EMG showed bilateral active denervation in the abdominal muscles depending on the T10, T11 and T12 roots, and in the paraspinal muscles at the T10 level. Soleus H-reflex latencies were prolonged. The patient was treated with ceftriaxone, 2 g/day for 14 days. Three months later the neurological examination was normal, although she still experienced intermittent moderate pain in the left lumboabdominal area. At that time, EMG showed reinnervation signs and a marked reduction of denervation signs in the abdominal muscles.

Discussion

Our three cases meet the criteria [2] for the diagnosis of neuroborreliosis:

1. All patients lived in an endemic area.
2. All presented with a painful polyradiculoneuritis demonstrated by clinical examination and EMG, as well as a CSF-proven lymphocytic meningitis.
3. All presented immunological evidence of exposure to *Borrelia burgdorferi,* with specific intrathecal antibody production.

In second-stage neuroborreliosis radiculoneuropathy starts, on average, 4–6 weeks (range 1–12) after the tick bite or the erythema migrans. Pain occurs first, usually in the limbs, with dysaesthesia, numbness or peripheral nerve paresis [3,4]. In our three cases the symptoms were prominent at uncommon thoracic (T10–T12) and lumbar (L1) levels.

Two patients also had a marked abdominal wall paresis. Common causes of abdominal distension (ileus, colonic obstruction, toxic megacolon, tumour, ascites) were easily ruled out. Four cases of abdominal wall weakness as the first presentation of neuroborreliosis have been reported [5–8]. All were preceded or accompanied by radicular pain or dysaesthesia. Three patients also had a facial palsy. Abdominal sensation was normal in two cases. As in our cases, the paresis was

bilateral in one patient. This man, devoted to body-building, noted progressive abdominal distension, the impossibility of performing his usual sit-up exercises, as in our first patient, and an increase in his trouser waist size from 34 to 42. In the three other reported cases the patients had unilateral abdominal distension that occurred only with the Valsalva manœuvre, and in one case while sitting up. Constipation was sometimes present but there was no other digestive symptom or sign. As in our patients, EMG showed active denervation in the abdominal muscles and CSF analysis revealed a lymphocytic meningitis with antibodies against *Borrelia burgdorferi*. In a recent German series of 90 cases of stage II neuroborreliosis, 26 patients presented a thoracic radiculoneuritis which was an isolated feature in only four [9]. The remainder had various additional neurological involvements, including cervical radiculoneuritis (9 cases), lumbosacral radiculoneuritis (18 cases) or cranial neuritis (10 cases). Eleven patients had abdominal wall weakness, which was bilateral in 10, the prominent feature in six and severe in only two. In the majority of these patients thoracic radiculoneuritis was located in the lower thoracic segments (T7–T12).

Thoracic radiculopathy rarely produces significant abdominal muscle paresis, probably because multi-segmental innervation is received from the intercostal nerves (T5–T12 roots) as well as the iliohypogastric and ilioinguinal nerves (L1 root). However, the absence of clinical abdominal muscle paresis does not mean that there is no motor involvement. Indeed, as in Case 3, EMG can demonstrate subclinical signs of denervation.

The differential diagnosis of abdominal wall paresis is summarised in Table 1. Abdominal wall paresis occurs in about half of the patients with diabetic thoracic polyradiculoneuropathy, usually with severe and long-lasting pain [10,11], but is occasionally painless [12]. Other diabetic complications, particularly polyneuropathy and retinopathy, are common. Cutaneous sensory abnormalities are nearly always present, sometimes only in the part of the dermatomal area involved with the pain. One of our patients did indeed have type II diabetes. It was mild, recent, well controlled and still uncomplicated. Herniation of a thoracic [13] or L1/L2 disc [14] can become symptomatic by unilateral paresis of the abdominal wall, but cannot explain in our patients either the multiple nerve root involvement or the lymphocytic meningitis. Moreover, MRI of the spine performed in Case 2 was unremarkable. Cutaneous herpes zoster in the area of the thoracic dermatomes is rarely associated with abdominal muscle paralysis [15]. However, systematic EMG studies of paraspinal muscles can show ipsilateral segmental motor involvement in 70% of patients who do not have a clinical weakness [16]. Radicular pain usually precedes zoster by 7–100 days [17]. Our patients had no vesicular rash during a follow-up period of 3 months and more. Zoster sine herpete, defined as dermatomal-distribution pain without rash, is possible, but this particular condition seems to be very unusual. The identification of another pathogenic agent likely to be responsible for the clinical picture in our patients and the good outcome with specific antibiotics discredit this hypothesis. Unilateral abdominal wall weakness has also been reported in association with syringomyelia [18]. MRI of the spinal cord excluded this diagnosis in the second patient and the CSF abnormalities also pointed to a different condition.

The involved roots in our cases led to misleading presenting symptoms, giving rise to the wrong initial diagnostic hypotheses. The first patient was suspected of a bowel disorder. The second was first investigated for urinary lithiasis. The third was referred to the gastroenterology department for a diagnostic work-up. Nevertheless, a careful enquiry (non-mechanical pain, exacerbation during the night or evening, absence of specific digestive symptoms except for constipation) and neurological examination (hypoaesthesia in radicular territories and abdominal muscle paresis) can suggest polyradiculoneuritis, leading to appropriate investigations, early diagnosis and treatment.

Table 1. Differential diagnosis of abdominal wall paresis

Aetiology	Diagnostic clues
Borrelia burgdorferi	Erythema migrans
	Tick bite
	Specific antibodies
	CSF analysis
Varicella zoster	Vesicular rash
	Specific antibodies
Diabetes	Long-standing history of diabetes
	Other diabetic complications
Disc herniation	Radicular pain increased by movement and effort
	Paravertebral muscle spasm
	Restriction of spinal movement
	MRI of the spine
Syringomyelia	MRI of the spine

References

1. Nadelman RB, Wormser GP. Lyme borreliosis. Lancet 1998;352:557–65.
2. Halperin JJ, Logigian EL, Finkel MF, Pearl RA. Practice parameters for the diagnosis of patients with nervous system Lyme borreliosis (Lyme disease). Quality Standards Subcommittee of the American Academy of Neurology. Neurology 1996;46:619–27.
3. Garcia-Monaco JC, Benach JL. Lyme neuroborreliosis. Ann Neurol 1995;37:691–702.
4. Kaiser R: Neuroborreliosis. J Neurol 1998;245:247–55.
5. Daffner KR, Saver JL, Biber MP. Lyme polyradiculoneuropathy presenting as increasing abdominal girth. Neurology 1990;40:373–5.
6. Krishnamurthy KB, Liu GT, Logigian EL. Acute Lyme neuropathy presenting with polyradicular pain, abdominal protrusion, and cranial neuropathy. Muscle Nerve 1993; 16:1261–4.
7. Vial C, Petiot P, Latombe D et al. Paralysie des muscles larges de l'abdomen due à une maladie de Lyme. Rev Neurol (Paris) 1993;149:810–12.
8. Roche C, Durieux A, Clavelou P. Pseudo-éventration révélant une maladie de Lyme. Ann Med Interne (Paris) 1997;148:188–90.

9. Pfadenhauer K, Schonsteiner T, Stohr M. Die thorakoabdominale manifestation der Lyme-neuroborreliose stadium II. Nervenartz 1998;69:296–9.
10. Kikta DG, Breuer AC, Wilbourn AJ. Thoracic root pain in diabetes: the spectrum of clinical and electromyographic findings. Ann Neurol 1982;11:80–5.
11. Longstreth GF. Diabetic thoracic polyradiculopathy: ten patients with abdominal pain. Am J Gastroenterol 1997;92:502–5.
12. Parry GJ, Floberg J. Diabetic truncal neuropathy presenting as abdominal hernia. Neurology 1989;39:1488–90.
13. Meyer F, Feldmann H, Toppich H, Celiker A. Unilaterale parese der bauchwandmuskulatur durch einen thorakalen bandscheibenvorfall. Zentralbl Neurochir 1991;52:137–9.
14. Billet FP, Ponssen H, Veenhuizen D. Unilateral paresis of the abdominal wall: a radicular syndrome caused by herniation of the L1–2 disc? J Neurol Neurosurg Psychiatry 1989;52:678.
15. Gottschau P, Trojaborg W. Abdominal muscle paralysis associated with herpes zoster. Acta Neurol Scand 1991;84:344–7.
16. Greenberg MK, McVey AL, Hayes T. Segmental motor involvement in herpes zoster: an EMG study. Neurology 1992;42:1122–3.
17. Gilden DH, Kleinschmidt-DeMasters BK, LaGuardia JJ, Mahalingam R, Cohrs RJ. Neurologic complications of the reactivation of varicella-zoster virus. N Engl J Med 2000;342:635–45.
18. Coleman RJ, Ingram DA. Abdominal wall weakness due to thoracic syringomyelia. Neurology 1991;41:1689–91.

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