Current Trends in Internal Medicine
Bruijn NRA, et al. Curr Trends Intern Med 8: 223.
www.doi.org/10.29011/2638-003X.100123
www.gavinpublishers.com

Case Report

An Unusual Presentation of Lyme disease In a Patient with Severe Abdominal Pain: A Case Report

N.R.A. Bruijn[1*], L. Slobbe[1,2], B. Özcan[1]

[1]Department of Internal Medicine, Erasmus MC, University Medical Center Rotterdam, The Netherlands

[2]Department of Medical Microbiology and Infectious Diseases, Erasmus MC, University Medical Center Rotterdam, The Netherlands

[*]**Corresponding author:** Noor Bruijn, Erasmus University Medical Center Rotterdam, PO Box 2040, 3000 CA Rotterdam, The Netherlands

Citation: Bruijn NRA, Slobbe L, Özcan B (2024) An Unusual Presentation of Lyme disease In a Patient with Severe Abdominal Pain: A Case Report. Intern Med 8: 223. DOI: 10.29011/2638-003X.100123

Received Date: 15 February 2024; **Accepted Date:** 22 February 2024; **Published Date:** 25 February 2024

Abstract

Objective: We present thoracic radiculopathy as the sole clinical sign of early disseminated neuroborreliosis.

Methods: We report a case of a 40-year-old female with panhypopituitarism and a permanent ventriculoperitoneal drain who experienced severe abdominal pain as a primary and isolated symptom of Lyme disease (LD).

Results: To our knowledge, thoracic radiculopathy as an isolated neurological manifestation of LD has been presented before only once. Radiculopathy is a well-known symptom of LD, but usually occurs in conjunction with additional neurological symptoms. The diagnosis was supported by a self-reported abdominal skin lesion consistent with erythema migrans and by additionally performed serological Borrelia tests.

Conclusion: Although rare, thoracic radiculopathy may be an isolated symptom of early neuroborreliosis. Alternative diagnoses have to be taken into account. As an additional learning point, doctors should be aware of the potential abrogation of the usual evolution of the antibody response due to adequate treatment of the infection.

Keywords: Lyme Disease; Thoracic radiculopathy; Lyme neuroborreliosis; Abdominal pain

Introduction

More than 200,000 patients are diagnosed with Lyme disease (LD) due to spirochetal infection with one of the pathogenic *Borrelia* species in Europe each year [1,2]. Since the clinical picture of LD varies, establishing a proper diagnosis in patients not exhibiting a classical disease presentation may be difficult. Here we present a patient with self-reported erythema migrans (EM) followed by disabling thoracic radiculopathy as an isolated neurological symptom due to early disseminated Lyme disease.

Case Report

We describe a 40-year-old female patient with severe abdominal pain due to thoracic radiculopathy as the initial presentation of early disseminated Lyme disease (LD). Her medical history comprised the removal of a craniopharyngioma followed by stereotactic radiotherapy in her teens which was complicated by an obstructive hydrocephalus for which a permanent ventriculoperitoneal (VP) drain was placed. For many years her clinical situation remained stable. Full hormonal supplementation was given in view of panhypopituitarism. During an earlier hospitalization due to COVID-19 in 2022, a CT scan coincidentally showed an asymptomatic right adnexal cyst with a diameter of 17 cm for which a wait-and-see policy was approved due to a lack of symptoms.

In August 2022, she was referred because of progressive and continuous abdominal pain over the past three weeks. Four days prior to this pain, she had noticed a circular red area with central clearing on the lateral right side of her abdomen with a diameter of approximately 15 cm. She described this skin lesion as itchy with a burning and stabbing sensation. Unfortunately, she did not take a picture of this skin lesion. On a website showing pictures of skin diseases, she had seen a picture of EM that looked very similar to her own skin defect. She lives in a forested region and takes

frequent walks. She did not detect ticks on her body. After four days, the rash disappeared but, in return, severe abdominal pain appeared, mainly located at the right lateral side of her abdomen and radiating backwards. She had no fever, nausea, diarrhea or obstipation. There were no pulmonary, cardiac or joint complaints. She had already doubled her hydrocortisone dose without any improvement. Findings upon physical examination were unremarkable apart from pain during palpation of the abdominal right side. Apart from low free thyroxine (FT4), laboratory blood tests showed no abnormalities. The infection parameters were low, with an EST of 19 mm/U and a CRP level of 15 mg/L, which was slightly elevated.

An abdominal CT scan showed nothing remarkable. There were neither signs of dilated bile ducts nor retroperitoneal lymphadenopathy, ascites, or bowel wall thickening. The adnexal cyst was unchanged, without signs of rupture or torsion. The tip of the VP drain was visible at the left side of her abdomen, with no fluid collection surrounding the tip. The drain tract was unremarkable. Her symptoms did not meet the diagnostic criteria for abdominal cutaneous nerve entrapment syndrome (ACNES) and were not consistent with bowel colic. Celiac disease had been previously excluded by serology. Based on the self-reported skin lesion, we considered EM followed by thoracic radiculopathy due to early disseminated LD, for which doxycycline was started.

In classical cases, EM is diagnosed solely on clinical grounds with no need for additional laboratory tests. However, as the patient's abdominal pain did not fit well within the classic presentation of LD, additional serological antibody-based tests against *Borrelia* were carried out. The routinely performed enzyme-linked immunosorbent assay (ELISA) test turned out to be positive, thus a western blot confirmation test was performed. Apart from two clearly positive IgM bands, indicating the presence of antibodies against the p18 and p41 antigens, the IgM immunoblot showed an indeterminate reaction to the OspC antigen. This combination almost met the criteria for a positive result. The IgG immunoblot assay was undoubtedly positive.

Despite antibiotic treatment, the disabling abdominal pain persisted for 8 weeks and then resolved. During additional clinical analysis, no alternative explanations for her symptoms were found. Follow-up serological testing showed a western blot test result similar to those of the initial tests, with no significant titer increase. In June 2023, she was still not experiencing abdominal pain or other neurological complaints.

Discussion

To the best of our knowledge, thoracic radiculopathy as an isolated neurological manifestation of LD has been presented before only once.

Since the clinical presentation of LD varies broadly, making a clear diagnosis can be challenging. The clinical manifestations of LD are divided into three recognized stages of disease: early localized, early disseminated, and late disease [3]. However, the clinical features of these stages may have a considerable degree of overlap [3]. Moreover, patients presenting with early disseminated or late disease manifestations may lack a history of prior symptoms that could be attributed to early disease [4]. Many signs and symptoms lack specificity and may just as well be caused by other conditions. In our patient, we excluded alternative explanations for her symptoms. She was treated with doxycycline due to the self-reported skin lesion, suspicious for EM, which is the presenting symptom in 80% in LD [3]. Early disseminated LD usually occurs within weeks to several months after EM, and may be the first manifestation of LD. In early disseminated LD, neurological involvement may occur in 3-15% of patients and includes lymphocytic meningitis, cranial nerve palsies, polyradiculitis, and peripheral neuropathy [3,5]. As for the abdominal pain, which in our patient lateralized to her right side and radiated to the right side of the back, we suspected thoracic radiculitis as the presenting symptom of early disseminated neuroborreliosis.

Although our patient's presentation was not typical, abdominal pain has previously been described as a manifestation of LD. In general, the symptoms of radiculitis are known to develop on average 4-6 weeks after the tick bite or EM [5]. Patients often describe the pain as burning or stabbing and is initially localized where the tick bite or EM has first been observed [5]. Radicular pain often peaks within hours or days, but can last for weeks to months [5]. Other neurological deficits often develop besides radicular pain [5]. Our patient experienced the disabling pain for eight weeks and it remained the only neurological disease manifestation. While the literature on thoracic radiculopathy as a manifestation of LD is scarce, a few recent case reports have described this symptom as the first clinical symptom of early disseminated neuroborreliosis with additional neurological symptoms, namely facial palsy, cranial neuropathy and ataxic gait [6-10]. In other case reports, abdominal wall weakness has been mentioned as an additional symptom for early disseminated neuroborreliosis [11-13]. One case report described abdominal pain as an isolated symptom, as in our patient [6].

In classical presentations of EM, the diagnosis of early localized LD should be made solely on clinical grounds. In this case, however, we decided to perform additional serological tests, as isolated and severe abdominal pain is not considered a classical sign of LD. Despite its limitations, serology is currently the main diagnostic laboratory approach available for LD [3]. Nevertheless, clinicians should bear in mind that patients with early localized disease may not develop a detectable antibody response until up to four weeks after infection [3,14]. Usually, a positive ELISA will be followed by western blot confirmation. These tests are usually both positive in patients with early disseminated LD, however, either one or even both tests can be negative [14]. In our patient, the primary assessment for antibody response was performed

while she was being treated with antibiotics. She already had been suffering from abdominal pain for four weeks. The initial ELISA screening assay was positive and the confirmatory IgM immunoblot showed an inconclusive test result, including two clearly positive bands and an indeterminate reaction to the OspC antigen. This may have been an indication of an early immune response. The IgG immunoblot also showed a clear positive test result. Three weeks later, repeated serological testing showed no significant increase in antibody titer which can be explained by abrogation of the antibody response. This may occur due to adequate antimicrobial treatment, which is in line with the results of a recent study [15]. Glatz et al. found that initially nonreactive patients, who were treated during early stages of disease, remained seronegative during one year of follow-up [15]. Within the seropositive patients in their study, they also observed no further antibody titer increase after antimicrobial therapy during one year of follow-up [15].

Conclusion

Thoracic radiculopathy may present as the primary and sole manifestation of early disseminated LD and we emphasize awareness for this presentation of LD. Although abdominal pain due to localized thoracic radiculopathy has been reported previously in other cases, care should also be taken to rule out alternative diagnoses. As a final point, although in classical cases of EM no further laboratory tests are recommended in clinical guidelines, serological tests may be of value in less typical disease presentations, as in our patient. We also note that adequate antibiotic treatment may hamper the continuation of the serological response, which has been reported before.

Declaration of interest: None

Informed consent: The patient consented to the use of her data for this case report

Declaration of the use of generative AI or AI-assisted technologies: None

Author's contribution

N.R.A. Bruijn: Wrote the orginal draft, conceptualized this manuscript, investigated literature, managed project administration, approved final version.

L. Slobbe: Conceptualized this manuscript, provided critical reviewing and editing, performed supervision, approved final version.

B. Özcan: Conceptualized this manuscript, provided critical reviewing and editing, performed supervision, managed project administration, approved final version.

References

1. Stanek G, Strle F (2018) Lyme borreliosis-from tick bite to diagnosis and treatment. FEMS Microbiol Rev 42: 233-58.
2. Marques AR, Strle F, Wormser GP (2021) Comparison of Lyme Disease in the United States and Europe. Emerg Infect Dis 27: 2017-24.
3. Schoen RT (2020) Challenges in the Diagnosis and Treatment of Lyme Disease. Curr Rheumatol Rep 22: 3.
4. Steere AC (2001) Lyme disease. N Engl J Med 345: 115-25.
5. Rauer S, Kastenbauer S, Hofmann H, Fingerle V, Huppertz HI, Hunfeld KP, et al. (2020) Guidelines for diagnosis and treatment in neurology - Lyme neuroborreliosis. Ger Med Sci 18: Doc03.
6. Stolk JM, van Nieuwkoop C, van der Voorn M, van Erp S, van Burgel ND (2018) Ticking off diagnoses of abdominal pain: early neuroborreliosis with radiculopathy. Neth J Med 76: 336-8.
7. Savasta S, Fiorito I, Foiadelli T, Pichiecchio A, Cambieri P, Mariani B, et al. (2020) Abdominal pain as first manifestation of lyme neuroborreliosis in children, case report and review of literature. Ital J Pediatr 46: 172.
8. Zulfiqar S, Qureshi A, Dande R, Puri C, Persaud K, Awasthi S (2021) The many manifestations of a single disease: neuroborreliosis. J Community Hosp Intern Med Perspect 11: 56-9.
9. Anas H, Jeffrey G (2017) Cervical Radiculopathy as the Initial Presentation for Lyme neuroborreliosis (P1.311). Neurology 88: P1.311.
10. Satinderpal S, Sayyeda Z, Irene C, Atef K, Jay V (2023) Syndrome of Inappropriate Antidiuretic Hormone Secretion - A Rare Initial Presentation of Lyme Neuroborreliosis (P9-10.005). Neurology 100: 4511.
11. Mormont E, Esselinckx W, De Ronde T, Hanson P, Deltombe T, Laloux P (2001) Abdominal wall weakness and lumboabdominal pain revealing neuroborreliosis: a report of three cases. Clin Rheumatol 20: 447-50.
12. Pfadenhauer K, Schönsteiner T, Stöhr M. (1998) [Thoraco-abdominal manifestation of stage II Lyme neuroborreliosis] Die thorakoabdominale Manifestation der Lyme-Neuroborreliose Stadium II. Nervenarzt 69: 296-9.
13. Krishnamurthy KB, Liu GT, Logigian EL (1993) Acute Lyme neuropathy presenting with polyradicular pain, abdominal protrusion, and cranial neuropathy. Muscle Nerve 16: 1261-4.
14. Rebman AW, Crowder LA, Kirkpatrick A, Aucott JN (2015) Characteristics of seroconversion and implications for diagnosis of post-treatment Lyme disease syndrome: acute and convalescent serology among a prospective cohort of early Lyme disease patients. Clin Rheumatol 34: 585-9.
15. Glatz M, Golestani M, Kerl H, Müllegger RR (2006) Clinical relevance of different IgG and IgM serum antibody responses to Borrelia burgdorferi after antibiotic therapy for erythema migrans: long-term follow-up study of 113 patients. Arch Dermatol 142: 862-8.