Case Report

Chronic Lyme disease presented with gastroparesis

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Abstract
We present a case of a 54-year-old White man who was admitted to our clinic for evaluation of gastroparesis. His gastroparesis was severe and unresponsive to previous treatments. Darkfield microscopy revealed the presence of spirochetes and corkscrew-shaped bacteria; although Lyme immunoglobulin M (IgM) and immunoglobulin G (IgG) Western Blot testings were negative. The patient was diagnosed with Chronic Lyme disease and recovered with antibiotherapy. We outline a rare case of dysmotility syndrome; a unique presentation of Chronic Lyme disease and emphasize the limitation of tools necessary in diagnosing Lyme disease.

Introduction
Chronic Lyme disease is an ongoing Borrelia infection that can involve any body system or tissue. Although subclinical hepatitis is the most common gastrointestinal manifestation of Lyme disease, gastroparesis has been rarely reported [1,2]. We present a patient with gastroparesis as an unusual presentation of chronic Lyme disease.

Case description
A 54-year-old White man was admitted to the hospital with a two-month history of postprandial bloating, nausea and weight loss. The medical history of the patient was significant for coronary artery disease and thymoma; which was cured 9 years ago. The patient was previously evaluated in two different hospitals before presenting to our clinic. Gastrointestinal scintigraphy revealed gastroparesis. The patient did not have a history of diabetes and his blood glucose level was normal. Therefore, diabetic gastroparesis was excluded as a possible diagnosis. Since idiopathic gastroparesis is the most common cause of gastroparesis [3], prokinetic treatment for idiopathic gastroparesis was initiated; however, symptoms did not subside. Next, enteral feeding was attempted. However, because the patient could not tolerate it, Total Parenteral Nutrition (TPN) was started.

On imaging screening, thoracic Computed Tomography (CT) followed by Positron Emission Tomography-Computed Tomography (PET-CT) revealed pleural malignancy. Pathology report of the pleural malignancy described relapse of thymoma, which was resected 9 years ago. Prior extensive workup including upper endoscopy, thoracic CT, contrast-enhanced abdominopelvic CT, abdominal ultrasound and PET-CT and laboratory results failed to demonstrate any other underlying significant findings. Due to concomitant thymoma, further investigation and treatment were initiated for the paraneoplastic syndrome and myasthenia gravis as potential underlying causes of gastroparesis. Acetylcholine receptor antibody, paraneoplastic autoantibody evaluation including anti-Hu, anti-Yo, anti-Ri, anti-Amphiphysin, anti-Tr, anti-PCA-2, anti-Ma, anti-CV2-1, anti-ANNA-3 and neostigmine test were all negative. In addition to metoclopramide, domperidone, erythromycin therapies, radiotherapy, pyridostigmine and prednisolone therapies were initiated, however, the symptoms did not diminish. An extensive literature search for gastroparesis was conducted via PubMed and Google Scholar. At the end of this whole process, neuroborreliosis was considered in the preliminary diagnosis due to the presence of similar cases in the literature; although very rare [1,2,4,5]. Dark field microscopy and Lyme IgM and IgG Western Blot (Euroimmun, Germany) testings were performed. The specific antibodies were negative, but the Dark field microscopy revealed the presence of spirochetes, corkscrew-shaped bacteria and cystic structures compatible with Borrelia species in the blood (Figure 1). Although cystic structures were compatible with Borrelia and there were no clinical signs of syphilis, we performed a further investigation to eliminate other spirochetal species such as treponema. Syphilis ELISA IgG (Vircell S.L., Granada, Spain), Elecsys® Syphilis (Roche Diagnostics, Mannheim, Germany) and TPHA (Spinreact, Girona, Spain) were used as treponemal tests in syphilis screening; VDRL (Spinreact, Girona, Spain) was used...
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as a non-treponemal test. Both of the results came negative, which aided in the exclusion of syphilis as a differential diagnosis. Since Lyme IgM and IgG Western Blot testings were negative, acute Lyme disease was excluded but the patient was diagnosed with chronic Borrelia infection. We initiated triple antibiotherapy - ceftriaxone and doxycycline; which are among the most effective antibiotherapies against Borrelia spirochetes and metronidazole which is effective against a cystic form of Borrelia. Radiotherapy, TPN and metoclopramide were continued. On the 12th day of treatment, the patient started oral feeding, after which TPN was stopped. He was discharged from the hospital on day 17 to continue the treatment at home. Upon discharge, the patient’s weight and body mass index were 46 kilograms (kgs) and 15.4 kg/m², respectively. At follow-up, after 3 months the patient’s weight increased to 50 kgs and a positive response to radiotherapy was observed in the PET-CT Scan.

Discussion

Both acute and chronic Lyme disease is caused by the bacteria Borrelia burgdorferi transmitted by some insects, most notably by Ixodes ticks [6]. It can elicit a multisystem inflammatory response, thus affecting any body system (or tissue), but generally involves the skin, musculoskeletal, cardiovascular and central nervous systems. Chronic Lyme disease can also cause gastrointestinal symptoms such as gastroparesis, as seen in this patient. Although the pathogenesis has not been fully elucidated, it is thought that dysmotility syndromes secondary to chronic Lyme disease may result from inflammatory neuropathy [1,2]. It is known that the genus of Borrelia can escape the immune system by producing biofilm, which makes the immune system unable to respond to the infectious agent. As a result, the molecular components of the immune system like acute phase reactants and immunoglobulins are mostly negative, as demonstrated in our case. However, the infection produces a wide range of signs and symptoms; namely rash, arthritis, carditis, neuropathy and rarely gastrointestinal dysmotility syndromes. In an article published in the Journal of Pediatric Gastroenterology and Nutrition, it has been stated that the diagnosis can be provided by polymerase chain reaction (PCR) analysis of gastrointestinal biopsies in the absence of blood test confirmation of Lyme [7]. Quantification of the infectious agent by dark-field microscopy can be useful when antibodies are negative in cases when Lyme disease is considered a part of the differential diagnosis. Lyme disease is rare in Turkey; however, the chronic form of Borrelia infection is probably more common than believed. Lyme disease should be kept in mind as a differential diagnosis for patients who present with gastroparesis unresponsive to treatment.

Conclusion

We outline a unique presentation of chronic Lyme disease which is a rare cause of dysmotility syndrome. Neuroborreliosis should be considered in the differential diagnosis of gastroparesis. Also, we emphasize the limitation of tools necessary in diagnosing Lyme disease.

Ethical disclosure: Written informed consent has been obtained from the patient.

Author contributions

Özlem ÖZYURT: Followed the patient during hospitalization.
➢ Collected the data
➢ Reviewed the literature
➢ Designed and drafted the manuscript.

Vedat Turhan: Followed the patient during hospitalization.
➢ Revised the manuscript
➢ Supervised the work.

References