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Case Report

Unusual Case of Leg Pain

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Abstract

Lyme disease is a tick-borne disease that may present with a wide range of musculoskeletal symptoms. Although arthritis is frequently seen as a complication of Lyme disease, myositis has rarely been described as a sequelae of the disease. This article presents a case of a 13-year old male who developed leg pain and swelling and was eventually found to have myositis on magnetic resonance imaging. An extensive workup only revealed positive Lyme serology. Subsequent treatment of his Lyme disease led to full resolution of symptoms. The presentation of myositis in Lyme disease is extremely rare, and only a few cases have been described in the literature. In addition to its rarity, this condition may also present a diagnostic challenge to healthcare providers.

Keywords: Lyme disease; Myositis; Pediatrics

Abbreviations

MRI: Magnetic Resonance Imaging

Introduction

Lyme disease is a tick-borne illness caused by the organism *Borrelia burgdorferi*. The disease may have a wide range of clinical manifestations affecting multiple systems of the body [1]. In particular, it can affect the musculoskeletal system and cause a number of early and late complications. The most well-known musculoskeletal complication is arthritis, which may be seen as a manifestation of late stage Lyme disease [2]. Patients may develop migratory pain in joints, muscles, and tendons early in the disease course. In the later stages of the disease, patients can develop joint swelling and pain affecting one or several of the large joints [3,4]. Although Lyme arthritis is well-described, the presentation of myositis in the setting of Lyme disease is extremely unique and has rarely been described in the literature.

Case Presentation

A previously healthy 13-year-old male presented with a three-day history of left leg pain and swelling. He stated that he had jumped off a six-foot playhouse and landed on his feet five days prior to presentation. He denied having any symptoms until two days later, when he developed severe pain and swelling in his left lower extremity. He had taken analgesics with minimal improvement in symptoms. He denied any fevers, chills, or night sweats. He denied any recent travel, exposures to infectious agents, or animal bites. He was initially sent to our facility due to concern for compartment syndrome.

On admission to our facility, he had significant pitting edema over his left calf, ankle, and foot with tenderness to palpation. He refused to bear weight in that limb due to pain. He had been afebrile prior to admission, but he had a fever of 102F after arrival. He did not have any rashes or signs of insect bites. Laboratory evaluation was significant for an erythrocyte sedimentation rate of 58mm/hr and C-reactive protein of 236.8mg/L (normal <8mg/L). His creatine kinase was within normal limits. X-rays of his left knee, tibia and fibula, ankle, and foot were negative for fractures. A bedside ultrasound showed a fluid collection in the left calf. Magnetic resonance imaging (MRI) of the left lower extremity was subsequently performed and revealed a fluid collection in the left calf area consistent with pyomyositis. He underwent irrigation and debridement of the left calf abscess and was started on empiric broad-spectrum antibiotics. All of his cultures came back negative. His clinical symptoms did not improve significantly even on antibiotics, and his inflammatory markers remained elevated. His hospital course was complicated by the development of left knee septic arthritis, which required a repeat irrigation and debridement. During his hospital stay, he underwent an extensive evaluation for infectious causes, including Tick-borne, Francisella, Z-fever, Bartonella, Lyme, and Febrile antibody panels. All of his workup was unremarkable except for Lyme western blot that was positive for IgG and IgM. He subsequently completed a four-week course of Ceftriaxone and eventually had full resolution of his symptoms and normalization of his inflammatory markers.

Discussion

Since the first case of Lyme myositis was reported in 1986, fewer than 30 cases have been described in the literature [5,6]. However it is truly unique in the pediatric population. Although there are case reports of orbital myositis secondary to Lyme disease in the pediatric population, based on our literature search, we have not come across any cases of non-orbital Lyme myositis in pediatric patients [7,8]. Interestingly, the majority of patients reported to have Lyme myositis came from Europe, with as few as six cases being reported in the United States. It is unclear what might cause this distribution of cases, although one hypothesis suggests that a greater diversity of pathogenic *Borrelia Burgdoferi* species in Europe may result in more varied clinical manifestations of Lyme disease than in the United States [9].

Most patients with Lyme myositis presented with local muscle involvement, though several case reports described patients who presented with diffuse muscle involvement [10,11]. Patients with peripheral Lyme myositis tended to present with myalgias, swelling, and weakness. Additionally, some patients had associated neurological complaints, including signs of neuropathy. Several patients also had dermatologic manifestations, including erythema migrans and acrodermatitis chronica atrophicans. In terms of laboratory findings,

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patients often had a normal or slightly elevated creatine kinase. However, inflammatory markers were frequently normal or only slightly elevated. Abnormalities were seen on electromyogram and nerve conduction studies in several patients, including decreased motor nerve conduction velocities [11].

Diagnosis generally was made by histopathologic findings on muscle biopsy. In most of the cases of Lyme myositis in the literature, the diagnosis was made by a combination of clinical symptoms as well as myopathological signs that suggested an inflammatory myopathy [12]. Additional evidence was provided by clinical history and serological evidence of Lyme disease. In cases of Lyme myositis, muscle biopsy findings showed mostly interstitial infiltrates consisting primarily of macrophages and lymphocytes which were often associated with muscle fiber degeneration. Spirochetes were also detected in affected muscle fibers on many of the biopsy specimens. A second method described in one case involved performing polymerase chain reaction testing on the fluid from affected musculature, such as synovial fluid from the knee [6]. Although it was effective in detecting *Borrelia burgdorferi*, it could also produce false positives since the high sensitivity meant that it could detect the organism in the blood as well [10]. Nevertheless, this method has potential to be used to help support a diagnosis of Lyme myositis. Interestingly, advanced imaging such as MRI has not been frequently used in the diagnostic process. At the time of our literature search, our patient is one of few cases of non-orbital Lyme myositis where MRI was used as part of the diagnostic modality [6]. Additionally, we made the diagnosis of myositis in this case without performing a muscle biopsy.

Successful treatment of Lyme disease with improvement in the symptoms of myositis helps to support the diagnosis of Lyme myositis. Treatment is typically with doxycycline (in older children and adults) or a cephalosporin. The duration of therapy has varied significantly in the literature, ranging from ten days to two months [6]. The majority of the reported patients had improvement in their musculoskeletal symptoms, although the time to clinical improvement varied from several days to more than a year. In one case series of eight patients, the duration of disease ranged from 5 days to 6 years [11]. Although our patient improved and had full clinical resolution of his symptoms after a prolonged course of antibiotics, there is not much data in the literature regarding long term outcomes of Lyme myositis.

Conclusion

Given the rarity of this condition, it may frequently be misdiagnosed by healthcare providers. Additionally, there remains little data on risk factors that are associated with the development of Lyme myositis. There is no data on whether pediatric patients have different outcomes compared to adult patients. We do know that compared to other patients in the literature, our patient had a more complex hospital course and experienced additional complications, namely the development of septic arthritis. It is also likely that Lyme myositis is under-diagnosed, as the symptoms of musculoskeletal pain may be seen in many patients with Lyme disease and also other conditions presenting with musculoskeletal pain. Therefore, Lyme myositis should be included in the differential for all patients who present with musculoskeletal complaints, positive Lyme serology, and an otherwise unremarkable workup. This should be considered even more for people who live in Lyme endemic areas. The pathophysiology of Lyme myositis is still unclear, and additional research is required to study the mechanism by which *Borrelia* infections cause this complication.

References

1. Biesiada G, Czepiel J, Leśniak M, Garlicki A, Mach T. Lyme disease: review. Arch Med Sci. 2012; 8: 978–982.
2. Arvikar S, Steere A. Diagnosis and Treatment of Lyme Arthritis. Infect Dis Clin North Am. 2015; 29: 269–280.
3. Steere A. Musculoskeletal manifestations of Lyme disease. Am J Med. 1995; 98: 44S-48S.
4. Bitar I, Lally E. Musculoskeletal manifestations of Lyme disease. Med Health R I. 2008; 91: 213-215.
5. Schmutzhard E, Willeit J, Gerstenbrand F. Meningopolyneuritis Bannwarth with focal nodular myositis. A new aspect in Lyme borreliosis. Klin Wochenschr. 1986; 64: 1204-1208.
6. Holmgren A, Matteson E. Lyme Myositis. Arthritis & Rheumatism. 2006; 54: 2697-2700.
7. Sauer A, Speeg-Schatz C, Hansmann Y. Two cases of orbital myositis as a rare feature of lyme borreliosis. Case Rep Infect Dis. 2011; 2011: 372470.
8. Seidenberg KB, Leib ML. Orbital myositis with Lyme disease. Am J Ophthalmol. 1990; 109: 13–16.
9. Cadavid D, Bai Y, Dail D, Hurd M, Narayan K, Hodzic E, et al. Infection and inflammation in skeletal muscle from nonhuman primates infected with different genospecies of the Lyme disease spirochete Borrelia burgdorferi. Infect Immun. 2003; 71: 7087–7098.
10. Atlas E, Novak SN, Duray PH, Steere AC. Lyme myositis: muscle invasion by Borrelia burgdorferi. Ann Intern Med. 1988; 109: 245–246.
11. Reimers CD, de Koning J, Neubert U, Preac-Mursic V, Koster JG, Müller-Felber W, et al. Borrelia burgdorferi myositis: report of eight patients. J Neurol. 1993; 240: 278–283.
12. Reimers CD, Pongratz DE, Neubert U, Pilz A, Hübner G, Naegele M, et al. Myositis caused by Borrelia burgdorferi: report of four cases. J Neurol Sci. 1989; 91: 215–226.

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