Spontaneous Rupture of Multiple Tendons in the Lower Extremity Following the Diagnosis of Lyme Disease

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Over a time frame of less than 1 year, a 23-year-old competitive horseback rider experienced a midsubstance tear of both the tibialis anterior and extensor hallucis longus tendons without inciting injury. It was after the second spontaneous tear that the patient's recent diagnosis of Lyme disease became the likely culprit. Often, patients with chronic Lyme disease present with an elaborate clinical picture that can mimic many more common diagnoses such as septic arthritis, transient synovitis, ligamentous sprain, and various other traumatic injuries. With the pathognomonic erythema migrans rash reported to be present less than 50% of the time in late-stage infections, the diagnosis of Lyme disease can often be difficult, with a high rate of underdiagnosis. It is important that Lyme disease be included in the differential diagnosis of spontaneous tendon pathology, especially for physicians practicing in highly endemic areas. The treatment is relatively simple and successful—especially for an acute infection—and it is important to initiate treatment promptly to prevent disability. (J Am Podiatr Med Assoc 109(6): 455-458, 2019)

Lyme disease was originally described in 1977 after an investigation of juvenile arthritis reported in children living in Lyme, Connecticut.[1] The Centers for Disease Control and Prevention began surveillance of Lyme disease in 1982 after an increasing incidence was observed. Occurring predominantly in the northeastern and north central regions of the United States, where the greatest prevalence of the *Ixodes scapularis* tick and the preferred host of the ticks (ie, deer) reside, Lyme disease has become the most common vector-borne disease in the United States.[2] Transmission of the disease occurs after a bite from an infected tick that has been attached for over 24 hours. The saliva of the tick with the circulating *Borrelia burgdorferi* spirochete is injected into the host's skin, bacterial growth occurs, and erythema migrans with the characteristic erythematous reaction occurs.[3,4] It is the hematogenous dissemination of the spirochete that causes multiple organ systems to be affected.[5] Infection has three phases: early, early disseminated, and late. Late infection is characterized by musculoskeletal complaints including arthritis months to years after initial symptoms, which has been known to occur in up to 10% of all patients regardless of appropriate treatment. Continued inflammation can develop in those patients who develop synovial autoimmunity to the original Lyme infection.[6]

Often, patients with chronic Lyme disease present with an elaborate clinical picture that can mimic many more common diagnoses such as septic arthritis, transient synovitis, ligamentous sprain, and various other traumatic injuries. Untreated Lyme disease can lead to various responses, including synovial hypertrophy; infiltration of mononuclear cells; vascular proliferation; and accumulation of cytokines, complement, immune complexes, and matrix metalloproteinases from chondrocytes and neutrophils in synovial fluid.[7,8] Large, weight-bearing, synovial joints are frequently affected. Symptoms can occur weeks to months after the inciting tick bite, and patients often present with intermittent episodes of joint pain and swelling. Late-stage manifestations of Lyme disease differ from case to case and range from arthritis to muscle aches, with tendon pathology infrequently reported. A bimodal age distribution of the ranges 5 to 9 years and 55 to 59 years is most common.[9,10] With the pathognomonic erythema migrans rash reported to be present less than 50% of the time in late-stage infections,[9,11] the diagnosis of Lyme disease can often be difficult, with a high rate of underdiagnosis.

Tendons, formed primarily from collagen and elastic fibers, support tissues and anchor skeletal

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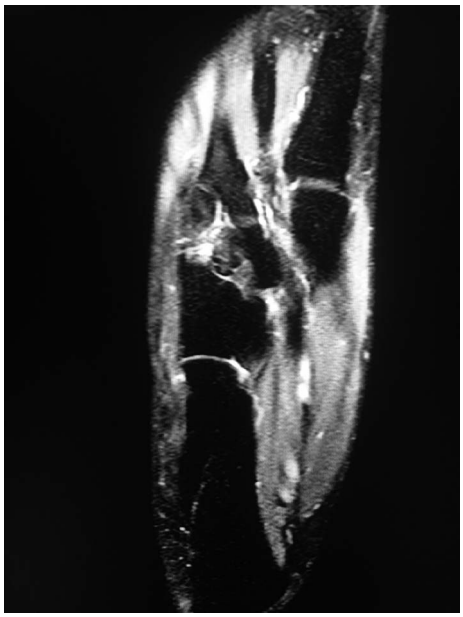


Figure 1. Axial T1-weighted magnetic resonance imaging scan showing rupture of the anterior tibial tendon at the insertion on the dorsal medial foot.

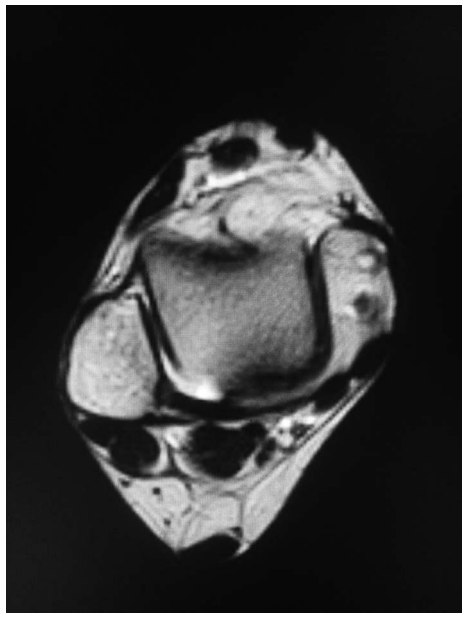


Figure 2. Axial T2-weighted magnetic resonance imaging scan showing extensor hallucis longus rupture just proximal to the extensor retinaculum.

muscle to the periosteum. Müller,[12] in his review of the histopathologic aspects of the damage of collagen elastic fibers caused by *B burgdorferi* in 2012, noted that *Borrelia* are capable of breaking down soluble and insoluble ground substance within the extracellular matrix by means of activation of metalloproteases. Collagen is dissolved and microcolonies of the bacteria can proliferate within the collagen fibers and inhibit regeneration promoted by fibronectin, subsequently delaying the healing process.[6,13,14] *Borrelia* interact with the glucosamine glycan hyaluronic acid, as it serves to regulate water binding and control permeability and diffusion. The binding of surface proteins to hyaluronic acid causes a reduction in pH, inactivation of proteoglycans, and a decrease in cross-linking, subsequently reducing viscosity. This ultimately leads to drying of the fibers within the tendon and to an increased risk of tendon rupture.[15,16]

In 2014, Miller et al[11] performed a single-center, retrospective medical record review of clinical data using the *International Classification of Diseases, Ninth Revision* code 088.81. All patients who had initially presented with lower-extremity pain, were not currently being treated for Lyme disease, did not have a history of Lyme disease, and did have a documented diagnosis of Lyme disease with laboratory records available were included in the study. In accordance with the Lyme disease two-tier testing guidelines, the first required test is the enzyme immunoassay or immunofluorescence assay. If the first test yields a positive or equivocal result, two options are available: 1) if the patient has had symptoms for 30 days or less, an immunoglobulin M (IgM) Western blot is performed; or 2) if the patient has had symptoms for more than 30 days, the IgG Western blot is performed. All of the patients were permanent residents (>3 years) of the metropolitan Philadelphia, Pennsylvania, area and symptoms had been present from 1 month to 1 year before the laboratory diagnosis. Only two patients (18.2%) recalled an inciting tick bite, and five (45.5%) described a rash consistent with erythema migrans. No patients underwent surgical intervention as a result of their Lyme-associated lower-extremity manifestations.[11]

In 2008, Pandya et al[17] published a case report of patellar tendon rupture as a manifestation of Lyme disease. Spontaneous midsubstance tearing was observed 13 months after trauma and DNA polymerase chain reaction (PCR) testing was positive for *B burgdorferi*. The tendon was repaired operatively, and a 3-week course of oral doxycycline was recommended by the infectious disease team. The 12-month follow-up revealed full return of strength and range of motion and negative Gram's stain, culture, and PCR testing, indicating eradication of joint infection.[3] There is very little literature for the specific treatment of tendon rupture

associated with Lyme disease specifically, but the tears must be treated like any other tendon rupture. Noninvasive techniques including both the Graston Technique (Indianapolis, Indiana) and the Active Release Technique (Atlas Injury Prevention Solutions, Grand Rapids, Michigan), followed by eccentric training and static stretching in combination with cryotherapy, have been shown to improve symptoms in patients with Achilles tendinopathy.[18] A patented form of soft-tissue mobilization, these techniques are theorized to reduce abnormal post-injury fibrous adhesions, to make scar tissue more mobile in subacute and chronic injury, and to facilitate healing in chronically degenerated soft tissues.[18] These techniques can be performed in a physical therapy setting.

Here, we present multiple spontaneous tendinous ruptures in a 23-year-old woman with Lyme disease. We informed our patient that data concerning her case would be submitted for publication, and written permission was obtained to publish this report. The patient is a competitive horseback rider born and raised in southern Florida. Over a time frame of less than 1 year, the patient experienced a midsubstance tear of both the tibialis anterior and extensor hallucis longus tendons without inciting injury. It was after the second spontaneous tear that the patient's recent diagnosis of Lyme disease became the likely culprit.

Discussion

The patient originally presented in the fall of 2016 with a chief complaint of tenderness at the insertion of the tibialis anterior tendon on the medial aspect of the right foot. She admitted to a possible sprain within the stirrup of her saddle while horseback riding 5 months previously, with intermittent pain failing to resolve. On physical examination, the patient was able to actively invert and dorsiflex the foot with a mild decrease in strength because of guarding. Mild effusion to the first metatarsal cuneiform joint was evident, with no ecchymosis. She was otherwise neurovascularly intact. Radiographs of the right foot and ankle were reviewed and were negative for any acute osseous or soft-tissue pathology. Clinical diagnosis of a strain with possible partial insertional tear of the tibialis anterior tendon was made and magnetic resonance imaging was performed. The scan was positive for spontaneous partial rupture of the tibialis anterior at its insertion. Conservative treatment of immobilization in a short-leg controlled ankle motion boot for 4 weeks was completed, and return to full activity was subsequently allowed. She was instructed to use wider stirrups and extra care when mounting and dismounting the horse to prevent future injury.

Four months later, she returned with insidious pain to the medial aspect of her right ankle which had, again, begun without injury. The only noted change from 4 months prior was her mention of a confirmed diagnosis of Lyme disease. On examination, the patient experienced tenderness over the medial ankle and dorsal foot with decreased active hallux dorsiflexion. There was no evidence of fracture or dislocation on plain radiographs. A repeated magnetic resonance imaging scan was ordered and showed extensive thickening of the extensor hallucis longus above, deep to, and slightly distal to the superior band of the extensor retinaculum, consistent with type 2 partial tearing. Conservative immobilization with a short leg walking cast for 4 weeks and transition into a controlled ankle motion boot for 2 weeks was completed, and symptoms resolved with full strength regained after a course of physical therapy.

Beginning in the summer of 2013, at the age of 20, the patient began feeling paranoid, with bouts of anxiety. She experienced episodes of fainting and inability to swallow occurring intermittently. Her constitutional symptoms progressed to hair loss and fatigue. Her symptoms were worked up by a rheumatologist, endocrinologist, and cardiologist over a period of 2 years, none of whom were able to provide a working diagnosis. After referral to a Lyme disease specialist in Orlando, Florida, it was discovered that her Lyme disease antibody and IgM blot were positive. The patient denied the observance of a tick or tick bite and could only associate her possible exposure to the many hours spent around the horses at various farms and stables. She was officially diagnosed with Lyme disease in 2014, and antibiotic therapy was initiated with doxycycline hyclate 100 mg for 18 months and Flagyl (Pfizer, New York, New York) 500 mg for 2 weeks.

Interestingly, the patient had previously suffered a right medial malleolar fracture to the affected extremity at the age of 17 years. Percutaneous reduction was performed with two 4.0-mm cancellous screws at the time of injury, and the hardware was subsequently removed 3 years later. All tendinous pathology occurred in her right lower leg and foot, which had previously sustained trauma 5 years before her diagnosis of Lyme disease. This is one of few reported cases of spontaneous rupture of multiple tendons in the lower extremity.

Conclusions

Our patient had a nontraumatic midsubstance tear of the flexor hallucis longus and an insertional tear of the tibialis anticus; therefore, we can postulate that the inflammation caused by the Lyme infection led to tendon degeneration and subsequent rupture. Consistent with late-phase Lyme disease, our patient's delay in development of midsubstance rupture occurred 2 years after her infection of Lyme disease. Her tendon ruptures were all noted within the same limb, which had previously undergone traumatic injury, as noted throughout the literature. Limitations of the report include the fact that PCR analysis was not completed on surrounding joints and the initiation of antibiotic therapy, as the patient had completed 18 months of suppression. This report documents a possible manifestation of Lyme disease in its late phase of an otherwise healthy young woman. It is important that Lyme disease be included in the differential diagnosis of spontaneous tendon pathology, especially for physicians practicing in highly endemic areas. The treatment is relatively simple and successful, especially in an acute infection, and it is important to initiate treatment to prevent disability.

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References

1. Bacon RM, Kugeler KJ, Mead PS, et al: Surveillance for Lyme disease—United States, 1992–2006. MMWR Surveill Summ **57:** 1, 2008.
2. Centers for Disease Control and Prevention: Lyme disease. Available at: http://www.cdc.gov/lyme/. Accessed May 1, 2018.
3. Pandya NK, Zgonis M, Ahn J, et al: Patellar tendon rupture as a manifestation of Lyme disease. Am J Orthop **37:** E167, 2008.
4. Steere AC: Lyme disease. N Engl J Med **345:** 115, 2001.
5. Schoen RT: Pathogenesis, diagnosis, manifestations, and treatment of Lyme disease. Curr Opin Rheumatol **3:** 610, 1991.
6. Coleman JL, Roemer EJ, Benach JL: Plasmin-coated *Borrelia burgdorferi* degrades soluble and insoluble components of the mammalian extracellular matrix. Infect Immun **67:** 3929, 1999.
7. Puius YA, Kalish RA: Lyme arthritis: pathogenesis, clinical presentation, and management. Infect Dis Clin North Am **22:** 289, 2008.
8. Steere AC, Duray PH, Butcher EC: Spirochetal antigens and lymphoid cell surface markers in Lyme synovitis: comparison with rheumatoid synovium and tonsillar lymphoid tissue. Arthritis Rheum **31:** 487, 1988.
9. Donta ST: Late and chronic Lyme disease. Med Clin North Am **86:** 341, 2002.
10. Smith BG, Cruz AI Jr, Milewski MD, et al: Lyme disease and the orthopaedic implications of Lyme arthritis. J Am Acad Orthop Surg **19:** 91, 2011.
11. Miller JR, Dunn KW, Braccia D, et al: Lyme disease manifestations in the foot and ankle: a retrospective case series. J Foot Ankle Surg **55:** 1241, 2016.
12. Müller KE: Damage of collagen and elastic fibres by *Borrelia burgdorferi*: known and new clinical and histopathological aspects. Open Neurol J **6:** 179, 2012.
13. Grab DJ, Kennedy R, Philipp MT: *Borrelia burgdorferi* posses a collagenolytic activity. FEMS Microbiol Lett **144:** 39, 1996.
14. Zambrano MC, Beklemisheva AA, Bryksin AV, et al: *Borrelia burgdorferi* binds to, invades and colonizes native type 1 collagen lattices. Infect Immun **72:** 3138, 2004.
15. Thein M, Bonde M, Bunikis I, et al: DipA, a pore-forming protein in outer membrane of lyme disease *Borrelia* exhibits specificity for permeation of dicarboxylates. PLoS One **7:** e36523, 2012. Published online May 10, 2012; doi: 10.1371/journal.pone.0036523.
16. Breves S: *Borreliose*. Colmberg: Medea-Verlag; 2007.
17. Piesman J, Maupin G, Campos EG, et al: Duration of adult female *Ixodes dammini* attachment and transmission of *Borrelia burgdorferi*, with description of a needle aspiration isolation method. J Infect Dis **163:** 895, 1991.
18. Miners AL, Bougie TL: Chronic Achilles tendinopathy: a case study of treatment incorporating active and passive tissue warm-up, Graston Technique, ART, eccentric exercise, and cryotherapy. J Can Chiropr Assoc **55:** 269, 2011.