Ten-year-old Omani Girl with Lyme Arthritis

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ABSTRACT

We report a 10-year-old girl who presented with acute arthritis of the left knee, which was confirmed as Lyme arthritis by serology and molecular assay. Careful history and examination were the keys to suspecting the diagnosis. The patient lived in the US for five years and her symptoms developed about a year after her return to Oman. To the best of our knowledge, this is the first case report of Lyme arthritis in Oman and the Arabian Gulf region. This case indicates the need to suspect Lyme disease in patients presenting with compatible symptoms and a history of recent travel to endemic regions.

Lyme disease is the most common tick-borne infection in the USA and Europe.[1,2] It is caused by the spirochete *Borrelia burgdorferi* and is primarily transmitted by *Ixodes* ticks.[1–3] It is endemic in North America, Europe, China, Japan, Mongolia, and states of the former Soviet Union.[3] The most common presentation of Lyme disease in children is erythema migrans, which occurs at the early stage, either localized at the site of the tick bite or disseminated throughout the body.[4] Constitutional symptoms, such as fever, malaise, and headache, may accompany the rash but it is not universal and generally is mild. If Lyme disease is not detected or treated at the early stages, it may progress into a late stage which manifests most commonly as arthritis and more rarely as isolated facial nerve palsy, Lyme carditis, or meningitis. The diagnosis of early Lyme disease is clinical, supported by a history of potential tick bite in a region where black-legged ticks are known or suspected to be present. All other clinical extracutaneous manifestations of possible Lyme disease should be supported by two-tiered serological testing, including an enzyme-linked immunosorbent assay screening test followed by a confirmatory Western blot test.[3] Lyme arthritis is inflammatory and usually affects a single large joint.[3] Arthritis can occur without a history of earlier stages of illness.[5] In fact, Lyme disease was first discovered after an epidemic of oligoarticular arthritis in Eastern Connecticut in the 1970s.[1] According to the Centers for Disease Control and Prevention, more than 30 000 cases of Lyme disease are reported annually in the USA. Up to a third of Lyme disease cases manifest as arthritis.[2] The absence of case reports of Lyme disease from the Arabian Gulf region including Oman suggests that it is not endemic in this region. To the best of our knowledge, this is the first case report of Lyme arthritis in Oman and its neighboring countries of the Gulf Cooperation Council.[6]

CASE REPORT

A 10-year-old healthy girl presented to Sultan Qaboos University Hospital's emergency department, Muscat, Oman in September 2020 with a one-week history of left knee pain, swelling, and limping. She first noted the swelling and pain acutely after waking up from sleep. The pain and swelling progressively worsened over the course of the week. Symptoms were exacerbated by motion and relieved by rest. She denied any history of fever or concurrent/preceding respiratory symptoms. She reported no preceding trauma or insect bites. No history of abdominal pain, change in bowel or urinary habits, skin rashes, or eye symptoms. No history of previous joint complaints or morning stiffness. She had no known sick contacts. She played with some farm animals a month before the presentation with no incidental bites or scratches reported. There was no history of ingestion of unpasteurized dairy products or undercooked meat. The patient did live with her family in the northern part of the state of New York, US, for five years when she used to participate in

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school camping trips and recalled receiving two tick bites about 2–3 years ago. She returned to Oman a year before developing the symptoms of arthritis.

Physical examination revealed a well-nourished, well-developed girl with normal vital signs. She had an antalgic gait but was able to bear weight partially on her left leg. Musculoskeletal examination revealed marked left knee swelling, mildly tender and warm to touch, extending 2 cm above and below the knee joint. There was mild erythema at the superolateral aspect of the joint. There was a partial restriction of active and passive range of motion. Examination of the other joints and systemic examination was otherwise normal.

Her laboratory investigations showed a normal leukocyte count and high C-reactive protein of 115 mg/L (normal range < 8 mg/L). Left-knee ultrasonography showed moderate effusion with synovial thickening. She underwent arthrocentesis in the emergency department which revealed a bloody tap with polymorphs of 95%. She was initially managed for acute septic arthritis with intravenous cefazolin. This was later changed to cefuroxime upon suspicion of Lyme arthritis, based on the revealed tick-bite exposure after her admission. Her knee mobility improved quickly after the arthrocentesis with a full normal range of movements attained, while the swelling and the erythema improved gradually afterward. The joint fluid culture did not grow any organisms. Interferon-gamma release assay for tuberculosis, as well as serologic tests for *Coxiella burnetii* and Brucella showed negative results. Magnetic resonance imaging of the knee with contrast was conducted and it showed a large knee joint effusion with diffuse thickening of the synovium with post-contrast enhancement but no evidence of osteomyelitis. Lyme disease enzyme-linked immunosorbent assay was reported to be strongly positive for both immunoglobulin M and immunoglobulin G. Polymerase chain reaction (PCR) identified *Borrelia burgdorferi* in the joint fluid. She was discharged home on day four of her hospital stay in good clinical condition and with downward-trending inflammatory markers. She made a full recovery by the end of the four-week treatment course.

DISCUSSION

Lyme disease is the most common tick-borne infection in the USA and Europe.[1,2] In North America, it is caused by the spirochete *B burgdorferi* and rarely by the recently discovered *Borrelia mayonii*. In Eurasia, *B burgdorferi, Borrelia afzelii,* and *Borrelia garinii* cause Lyme disease.[3]

Lyme disease occurs in three stages: early localized, early disseminated, and late disseminated disease.[3] Lyme arthritis is the most common manifestation of the late stage. The stages can overlap, and the late stage might present without noticeable early stage manifestation, as in the current case.[2] It can take months from the tick bite to the appearance of late manifestations like arthritis. Our patient gave a history of tick bites at least a year prior to her current presentation.

As Lyme disease cases had not been previously reported in Oman, Lyme arthritis was not initially entertained as a cause for our patient's septic arthritis. It was careful history and examination and consultation with infectious disease experts that led to the diagnosis. Indeed, a history of plausible geographic exposure to a tick bite is crucial for the early recognition and diagnosis of Lyme arthritis.[5]

Lyme arthritis manifests as monoarthritis or oligoarthritis, involving primarily large joints, especially the knees.[2,3] Lyme arthritis may be difficult to distinguish from septic arthritis, although distinguishing characteristics exist.[2,7] Table 1 summarizes the clinical differences between Lyme and septic arthritis. Our patient had a large knee

Table 1: Difference in clinical signs between Lyme and septic arthritis in children.

Clinical signs	Lyme arthritis	Septic arthritis
Fever	Uncommon	Common
Joint effusion	Usually large	Varies from small to large
Tenderness	Mild or absent	Severe
Range of motion	Patient can ambulate with some limitations	Patient may refuse to ambulate with significant restriction of movement
Erythema	Uncommon	Common

effusion with only mild pain, no fever, and only partial restriction in mobility. Furthermore, her peripheral-blood leukocyte count was normal. As such, her clinical findings were more suggestive of Lyme arthritis than acute septic arthritis.

Patients with Lyme disease may remain seropositive for years which may make the diagnosis challenging.[7] An initial positive serologic assay should be confirmed by a second-tier serologic test to increase specificity.[3] In our case, a second-tier test was not available, so we sought confirmation by PCR of the synovial fluid. Testing synovial fluid for Lyme disease PCR is still controversial due to the lack of Food and Drug Administration-standardized testing procedures and the potential for nonspecific results.[5]

Seronegative Lyme arthritis cases with detectable *Borrelia burgdorferi* DNA in synovial fluid or tissue have been documented in the literature.[8] Despite its limitations, PCR testing of the synovial fluid is generally considered to be informative in making the diagnosis of Lyme arthritis.[3] The Lyme disease review panel of the Infectious Diseases Society of America has recommended adding PCR to Lyme disease guidelines.[9] In our case, the combination of a suggestive clinical presentation, exposure to tick bites in an endemic region, positive serologic results, and positive PCR from the joint fluid left no doubt about the diagnosis.

Lyme arthritis in children is usually treated with doxycycline, amoxicillin, or cefuroxime for 28 days.[3,4,10] Nine out of 10 of patients respond well to antibiotic therapy.[5,11] Our patient made a full recovery with four weeks of oral cefuroxime.

CONCLUSION

Travel history is crucial in the early recognition and identification of infections not commonly seen in Oman. Acute onset monoarthritis with significant swelling that is out of proportion to pain, along with a history of travel to a Lyme-endemic area should raise the suspicion of Lyme arthritis.

Disclosure

The authors declared no conflicts of interest. Written consent was obtained from the patient's father.

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