Ten-year-old Omani Girl with Lyme Arthritis

Tahani Al Mughaizwi[1], Hatem Al Rawahi[2], Nagi Elamin[2], Zaid Al Hinai[2], Zakariya Al Muharrmi and Laila S Al Yazidi[2]*

[1]Oman Medical Specialty Board. Muscat, Oman

[2]Child health department, Sultan Qaboos University Hospital, Muscat, Oman

[3]Microbiology Department, Sultan Qaboos University Hospital, Muscat, Oman

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***Corresponding author:** lailay@squ.edu.om

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Abstract

Lyme disease may present as Lyme arthritis, the most common late manifestation of the disease. Lyme disease is endemic in North America, Europe, Russia and North East Asia. The available literature regarding the Middle East region is scarce and does not suggest that Lyme disease is endemic to Oman or neighboring countries. Here we report a 10-year-old healthy girl who presented with acute arthritis of the left knee, and was confirmed to have Lyme arthritis by serology and molecular assay. Careful history and examination were the key to suspecting the diagnosis. Our patient lived in the United States for 5 years and her symptoms developed about a year after return to Oman. Although her knee swelling was marked, her pain was mild, which was not typical of usual cases of acute septic arthritis. 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 recent travel to endemic regions.

Keywords: Lyme, arthritis, travel, children, endemic, tick.

Introduction

Lyme disease is the most common tick-borne infection in the United States and Europe.[1,2] It is caused by the spirochete *Borrelia burgdorferi* and is primarily transmitted by *Ixodes* ticks.[1,2,3] 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 as isolated facial nerve palsy, arthritis (most commonly), heart block (or carditis) or meningitis. The diagnosis of early Lyme disease is clinical, supported by a history of potential tick bite in an area where it is known or suspected that black-legged ticks have been established. All other clinical extracutaneous manifestations of possible Lyme disease should be supported by two-tiered serological testing, including an ELISA screening test followed by a confirmatory Western blot test.[3] Lyme arthritis is inflammatory, usually affect 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 (CDC), more than 30,000 cases of Lyme disease are reported annually in the United States. Up to a third of Lyme disease cases manifest as arthritis.[2] The available literature suggests that Lyme disease is likely not endemic in Oman. To the best of our knowledge, this is the first case report of Lyme arthritis in Oman and the countries of the Gulf Cooperation Council (GCC).[6]

Case report

We report a 10-year-old healthy girl who 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 her presentation with no incidental bites or scratches reported. There was no history of ingestion of unpasteurized dairy products or under-cooked meat. The patient did live with her family in the northern part of New York state, United States, for 5 years. She returned to Oman a year before developing the symptoms of arthritis. During her stay in the United States, she did go on school camping trips and recalled two tick bites about 2 -3 years ago.

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, extending 2 cm above and below the knee joint. The knee joint was only mildly tender and warm to touch. There was mild erythema at the superolateral aspect of the joint. There was 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 < 8mg/l). Left-knee ultrasonography showed moderate effusion with synovial thickening. She underwent arthrocentesis in the emergency department which revealed 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 reveled tick-bite exposure after her admission. Her knee mobility improved quickly after the arthrocentesis with full normal range of movements attained, while the swelling and the erythema improved gradually afterwards. 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. MRI 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 immunosorbent assay (EIA) was reported to be strongly positive for both IgM and IgG. Polymerase chain reaction to identify *Borrelia burgdorferi* in the joint fluid resulted as positive. She was discharged home on day 4 of her hospital stay in a very good clinical condition and with downward-trending inflammatory markers. She made a full recovery by the end of the 4-week treatment course.

Discussion

Lyme disease is the most common tick-borne infection in the United States 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 a noticeable early disease.[2] The period from the tick bite to the appearance of late manifestations, like arthritis, can be months. Our patient gave a history of tick bites at least a year prior to her current presentation.

There were no previous reports of Lyme disease cases in Oman by the time this case was reported. Lyme arthritis was not initially entertained as a cause for our patient's septic arthritis. However, careful history and examination, as well as consultation with infectious disease experts led to the diagnosis. Indeed, a history of plausible geographic exposure to a tick bite is very crucial for the early recognition and diagnosis of Lyme arthritis.[5]

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

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

Clinical signs	**Lyme arthritis**	**Septic arthritis**
Fever	Uncommon	Very common
Joint effusion	Usually large	Varies from mild to large
Tenderness	Mild or absent	Severe

Range of motion	Can ambulate with some limitation	Refuse to ambulate with significant restriction of movement
Erythema	Uncommon	Very common

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 FDA-standardized testing procedures and the potential for nonspecific results. [5]

Seronegative Lyme arthritis 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 (IDSA) have 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.[4,3, 10] 90% of patients with Lyme arthritis 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 early recognition and identification of some 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 for Lyme arthritis.

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