CASE REPORT

Unusual case of prosthetic joint infection caused by *Francisella Tularensis*

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SUMMARY

Tularaemia is a zoonotic infection caused by *Francisella tularensis*. Ulceroglandular, glandular, oculoglandular, pharyngeal, typhoidal and pneumonic types are the different types of the disease. Infection of prosthetic joints occurs at an exceedingly uncommon rate. We report a case of prosthetic joint infection involving the hip with *F. tularensis,* which to the best of our knowledge after a thorough literature review is the second of its kind.

BACKGROUND

Infection of prosthetic joints occurs at an exceedingly uncommon rate. The incidence has reduced to about 1%–2% depending on the type and site of prosthesis and if it is a primary or a revision procedure.[1] Zoonotic organisms are extremely rarely implicated in prosthetic joint infections. *Brucella melitensis* osteomyelitis involving an extra-articular bone implant and chronic Brucella prosthetic valve endocarditis has been described.[2] *Francisella tularensis* causes the disease commonly known as tularaemia also known as rabbit fever or deer fly fever. *F. tularensis* is not a common aetiology in prosthetic joint infections, however one case of ventriculoperitoneal shunt infection subsequent to rabbit exposure has been reported.[3] We report another case of prosthetic joint infection involving the hip with *F. tularensis,* which to the best of our knowledge is the second of its kind.

CASE PRESENTATION

A 77-year-old man with a medical history of right-sided total hip replacement (THR) done 25 years ago for osteoarthritis, presented with chief complaints of right-sided hip pain for about 7 days after a recent revision of his right THR. He had a significant history of being a hunter 50 years ago, but denies recalling any recent exposure to animals or tick bite. He denied having any pets at home. On initial presentation, he had severe right-sided hip pain worse with ambulation along with popping sensation and swelling around the joint. He then underwent a revision of his THR and was discharged home. Two intra-operative cultures were done from the joint, which did not reveal any infection. One week later he returned to the emergency room (ER) with worsening right-sided hip pain. On examination, he had a temperature of 100.6 °F (38.1°C). Range of movement on the right hip was limited due to the pain. No lymphadenopathy was noted. There was a bullous lesion noted on the shin, which, as per patient was present for almost a year associated with itching at times. The hip joint incision site showed no redness or drainage. Associated symptoms of lymph node swelling, difficulty breathing, eye irritation, sore throat, cough and chest pain were negative.

INVESTIGATIONS

Laboratory investigations showed a white cell count of 8400 cells/mL (4000−11 000 cells/mL) with a neutrophil count of 5600 (1.6–7.7×10^3/μL). Multiple complete blood counts during the patient's stay showed a white cell count within normal limits. The erythrocyte sedimentation rate (ESR) was 96 mm/h (0–15 mm/h) and C-reactive protein (CRP) was 16.20 mg/dL (0.0–0.29 mg/dL). X-ray of the right hip showed the hip prosthesis in anatomical alignment without any effusion or dislocation of the prosthesis. He was re-admitted to the hospital and an arthrocentesis was done which showed wet grey colonies on a culture medium using Vitek technology with no growth on MacConkey agar. Gram stain showed gram-negative bacilli and biochemical testing showed coagulase-negative bacilli with likely suspicion of Francisella organism at the hospital laboratory. To confirm the diagnosis, the sample was sent to a referral laboratory where using the MALDI system the organism was confirmed to be *F. tularensis*. The hospital laboratory was notified of this result from the referral laboratory. A repeat arthrocentesis was performed at the same hospital given the patient's rising ESR and CRP, which reconfirmed the above organism. All laboratory testings were performed using appropriate methodology for culture. As the joint fluid was available for culture, serological testing was deferred at that point. He was not given antibiotics during this period. To further confirm the organism, the aspirate was sent to referral laboratory at Illinois Department of Public Health laboratory, which showed growth of the same organism.

TREATMENT

The patient was started on oral doxycycline 100 mg two times per day. He followed up with infectious disease specialist and at the orthopaedic clinic in 1, 6 and 12 months after starting treatment.

OUTCOME AND FOLLOW-UP

His hip pain and bullous skin lesion had resolved completely. The hip joint incision site was well

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healed. The patient was clinically doing well on repeated follow-up.

DISCUSSION

Tularaemia is a zoonotic infection caused by *F. tularensis*. It is one of the most virulent species found almost exclusively in North America especially in the South Central USA, the Pacific Northwest and parts of Massachusetts.[4 5] It was much more common in the early part of the 20th century than it is now. As per the detailed literature review European countries had variable distribution of the disease with lower incidence reported in countries of Belgium, Bosnia, Italy, Germany and Switzerland in contrast to higher reported cases from Sweden, Norway, Bulgaria and Czech republic. In the USA, about 1208 cases of tularaemia were reported via National Notifiable Diseases Surveillance System during 2001–2010. It is found more commonly in men. Tularaemia predominantly is more prominent in the months of May through September, however it may occur at any time of the year.[5] *F. tularensis* is an aerobic and fastidious gram-negative bacterium which tends to exhibit bipolar staining with Gram stain or Giemsa stain methods.[4] *F. tularensis* and *F. holarctica,* commonly involved in human infections, are transmitted through invertebrate vectors like the dog tick (*Dermacentor variabilis*), the wood tick (*Dermacentor andersoni*) and the lone star tick (*Amblyomma americanum*). Deer flies have been shown to transmit tularaemia in the western USA. Transmission of the bacterium may occur by a direct contact with the skin or inhalation of aerosols or dust.[4 5] Clinical manifestations could range from asymptomatic illness to septic shock and death depending on the portal of entry. Incubation period is usually from 3 to 5 days but can range from 1 to 21 days.[4] Different forms of this disease are ulceroglandular, glandular, oculoglandular, pharyngeal, typhoidal and pneumonic types. All forms are accompanied with high grade of fever and lymph node swelling.[4] A confirmed case of tularaemia is defined when there is either a fourfold or greater change in serum antibody titre to *F. tularensis* antigen or isolation of *F. tularensis* from a clinical specimen. A probable case is defined with either a single elevated antibody titre to *F. tularensis* antigen or detection of *F. tularensis* in a clinical specimen by a fluorescent assay.[4] Tularaemia is a rare disease and diagnosis is difficult and can be mistaken for other illnesses. Blood tests and cultures can help diagnose the infection. A presumptive diagnosis of tularaemia may be made through testing of specimens using direct fluorescent antibody, immunohistochemical staining or PCR. Serological testing showing a fourfold change in antibody titres can be used to establish the diagnosis.[4 5] Antibiotics used to treat tularaemia include streptomycin, gentamicin, doxycycline and ciprofloxacin for a period of 7–14 days.[4] Avoiding exposure to the organism is the best prevention of tularaemia. Using insect repellent, wearing long sleeves/pants, removing attached ticks promptly with tweezers, avoiding contact with sick or dead animals and using gloves when handling animals, especially rabbits, muskrats, prairie dogs and other rodents, are some of the preventive measures.[4] Cases of laboratory-acquired infections have also been reported given *F. tularensis* being a highly infectious organism. Post exposure prophylaxis options for workers include doxycycline or ciprofloxacin for a total of 14 days. All personnel advised to watch for symptoms like fevers.[4 5] Vaccination for tularaemia is not available in the USA, nor is it useful in the management of ill patients.[4] Besides drainage of the fluid accumulated around the prosthetic joint as a part of diagnosis, the mainstay of treatment of any prosthetic joint infection is removal of the joint. There are no defined guidelines about the duration of antibiotic treatment of prosthetic joint infection with *F. tularensis*. Our patient was treated with oral doxycycline for a prolonged period of time of up to a year as he refused surgery. The role of chronic antibiotic use in such patients is not well defined but our patient has been clinically stable and has not had any further symptoms from the infection. To our knowledge, this is the second documented occurrence of *F. tularensis* prosthetic joint infection. It prompts us to elicit complete history to diagnose this unusual infection involving prosthetic joints.

Learning points

- This case will help clinicians to broaden their differential diagnosis while evaluating patients of hip joint pain and infection.
- It also prompts physicians to take a more complete history in order to formulate a complete diagnosis.
- Some level of suspicion should be kept for *F. tularensis* infection in patients presenting with bullous skin rash with joint infections.

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Competing interests None declared.

Patient consent Obtained.

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