Case Report: A Cluster of Three Leptospirosis Cases in a New York City Abattoir and an Unusual Complication in the Index Case

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Abstract. We report the case of a 46-year-old male abattoir worker who developed myalgias, shortness of breath, and irritability 2 weeks after sustaining a laceration to the hand with a knife at work. During his hospital evaluation for septic shock he was noted to be febrile, hypotensive, profoundly jaundiced with aseptic meningitis, and renal failure, and was diagnosed with *Leptospira interrogans* infection confirmed by serum and urine polymerase chain reaction. After standard antibiotic therapy and recovery from severe clinical illness, he developed unilateral orchitis with pyuria secondary to leptospirosis, a well-established complication in the veterinary literature, but of which we offer the first report in humans in the English literature. The case presented was also the index case that uncovered a cluster outbreak of leptospirosis in New York City during the winter of 2016–2017, involving a total of three patients who lived or worked within a block of the abattoir. Two patients survived whereas the third died of pulmonary hemorrhage shortly after seeking medical care.

A previously healthy 46 year-old man was admitted to the medical intensive care unit (ICU) at our hospital in December of 2016 with rapidly progressive multiorgan failure. He had been in good health until 8 days prior to presentation (PTA) when he developed watery diarrhea. Six days PTA, he noted bilateral thigh pain that was severe enough to limit his ability to walk or bear weight. Three days PTA he developed fevers with shivering chills, and 1 day PTA cough, bloody sputum, and shortness of breath which prompted him to seek medical help. In our emergency room his clinical status deteriorated rapidly, requiring the initiation of vasopressor support within the first 2 hours after presentation. Empiric antibiotics were started for presumed septic shock.

On review of systems, he reported a persistent headache as well as dizziness, tinnitus, and difficulty in sleeping for several days. He denied neck stiffness, visual changes, coryza, conjunctivitis, and rash. His spouse described him as anxious and increasingly irritable since the first onset of symptoms. He had no past medical or surgical history and took no regular medications. He had been using ibuprofen as needed for muscle pain since the onset of his present illness.

He was originally from the city of Puebla, Mexico but had immigrated to the United States 30 years prior. He did not use tobacco, alcohol, or illicit drugs. He owned no pets but worked as a slaughterer at an abattoir in the Bronx, New York where he had close contact with chickens, goats, cows, and pigs. To his knowledge, none of the animals had been sick; however, the patient's family reported that a fellow co-worker had died unexpectedly 2 weeks before after the sudden onset of hemoptysis.

On the morning after admission the patient was evaluated by the infectious diseases consultation service. He appeared acutely ill with a temperature of 39.6°C, pulse 145 beats/minute, respiratory rate of 18 breaths/minute, blood pressure 90/50 mmHg, and O₂ sat 92% on 4 L/minute of supplemental oxygen. He was awake, alert, and oriented to person and place. There was mild neck rigidity. His oral mucosa was dry and spattered with dry blood, and his skin and sclerae were

jaundiced. There was neither a conjunctival injection nor a skin rash. Scattered rales were heard throughout the lungs. There were no cranial nerve deficits although he complained of bilateral tinnitus. He had marked tenderness to palpation over calf and thigh-muscles bilaterally, and he was unable to move his lower extremities because of pain. There was a healing, deep, 3 cm laceration near the base of the left thumb from an injury sustained with a sharp blade while at work.

Initial laboratory studies were remarkable for a total white blood cell count of 12,000 cells/ μ L (91% neutrophils and 8% bands), a hemoglobin of 9.2 g/dL, and a total platelet count of 110×10^9 cells/L. Serum creatinine was 6.5 mg/dL, and venous bicarbonate 12 mEq/L. Aspartate transaminase 133 U/L, alanine transaminase 87 U/L, total bilirubin 8.4 mg/dL (peak of 14.1), direct bilirubin 5.9 mg/dL, and creatine phosphokinase 2348 U/L. Cerebrospinal fluid (CSF) analysis revealed 11 WBC/mL and 620 RBC/mL with 51 mg/dL of protein and 63 mg/dL of glucose. Urinalysis revealed large protein but no cells or casts. Human immunodeficiency virus antibodies were negative.

A presumptive diagnosis of severe leptospirosis was made based on history and exam. He was treated with targeted antimicrobial therapy with ceftriaxone and doxycycline. He was also started on hemodialysis for acute renal failure with acidosis. His respiratory status continued to decline and he required endotracheal intubation within 6 hours of admission. Chest X-rays progressed from mild bibasilar atelectasis (Figure 1A) to diffuse bilateral patchy opacities in that timeframe (Figure 1B), consistent with the acute respiratory distress syndrome (ARDS).

Given the degree of pulmonary involvement, the possibility of a Jarisch–Herxheimer reaction was considered. Based on reports of improved outcomes with high dose corticosteroids, hydrocortisone 100 mg every 8 hours for 3 days were administered.¹

Blood, urine, and CSF bacterial cultures revealed no growth. *Leptospira* DNA was detected by looking at LipL32 DNA by real-time polymerase chain reaction (PCR) in both urine and serum obtained on the second day after admission. Initial leptospirosis antibodies were negative but the presence of antileptospira antibodies at a titer of 1:3,200, predominantly reacting against serogroup Icterohaemorrhagiae, was confirmed in convalescent serum 4 weeks after initial presentation.

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FIGURE 1. Rapidly progressive, bilateral pulmonary opacities.

The patient was successfully extubated on hospital day 3, his fevers subsided, and the hyperbilirubinemia and leukocytosis resolved after a 7 day course of antibiotics. He required renal replacement therapy for an additional 3 weeks with complete recovery of renal function at 1-month follow-up.

Given the initial suspicion for the diagnosis of Leptospirosis and the concern for other possible cases in the city given the unexpected death of the patient's co-worker, the New York State Department of Health was contacted, and an investigation was launched into the abattoir where they both had worked. The co-worker was reported to be an otherwise healthy African American man in his early 30's who died of pulmonary hemorrhage shortly after arriving in a Bronx emergency room. His death had been initially attributed to heart failure, and leptospirosis infection was diagnosed postmortem by the city's medical examiner. A third case of leptospirosis was diagnosed 6 weeks later in a 44-year-old Hispanic man who lived in a basement apartment on the same block as the abattoir. He presented with fevers and hemoptysis and also developed acute multiorgan failure. He recovered fully after 2 weeks in an ICU at a hospital in the Bronx and had no known further complications. All three infections were acquired in the same block of the Melrose neighborhood in the Bronx, between mid-December 2016 and February 2017. All cases were confirmed by positive Leptospirosis PCR (LipL32) from serum samples sent to the Centers for Disease Control and Prevention (New York City Department of Health and Mental Hygiene [NYC DOH], March 2017, personal communication), and they represent the first cluster of leptospirosis infections reported to have occurred in New York City.² An on-site visit by the DOH to the abattoir uncovered a rodent infestation in that building and several adjacent ones.

Six weeks after the initial presentation to the hospital, the patient we took care of was seen for follow-up where he complained of several days of testicular pain and swelling. Exam revealed a temperature of 38.4°C and a moderately swollen and tender left testicle consistent with unilateral orchitis. There was no inguinal lymphadenopathy, no urethral discharge and no induration of the epididymis. Urinalysis revealed two + leukocytes without bacteria, and urine culture remained negative; gonorrhea and *Chlamydia* were ruled out by nucleic acid amplification test. Symptoms and pyuria resolved a week later without further antibiotics.

This case of severe leptospirosis in an abattoir worker was likely acquired following a deep tissue laceration with a knife

that was presumably contaminated with urine of a leptospira-infected animal. Classic signs and symptoms including fever, myalgias, diarrhea, jaundice, aseptic meningitis, oliguric renal failure and ARDS were initially present.³ These resolved after a course of antibiotics and steroids but were followed 6 weeks later by the development of orchitis, an uncommonly described complication of infection with *Leptospira*.

To our knowledge, a single case of orchitis in a human after Leptospirosis infection has been reported in the literature. The case, published in 1961, details the development of fever and bilateral orchitis at day 24 after a patient had contracted an infection with *Leptospira ballum* after a rat bite in a research laboratory in Switzerland. Proteinuria, leukocytes, and erythrocytes were documented in the patient's urine with a spontaneous resolution of signs and symptoms 3 days later.⁴

Two overlapping phases of leptospirosis infection are known to occur. The first, characterized by multiorgan involvement, and thought to result from high levels of leptospiremia, is followed a short time later by a phase of leptospiruria after the spirochetes have been cleared from the blood.⁵ The latter phase is thought to persist for 4–8 weeks after recovery from acute infection and may be related to the development of orchitis as occurred in this case.

Orchitis is well documented in the veterinary literature as a common complication of Leptospirosis in male cattle.⁶ Presence of leptospermia in experimentally inoculated rams was demonstrated in a 1964 experiment which showed sperm infection at the level of the epididymis, ruling out the possibility of contamination of ejaculated sperm from a urinary source.⁷ Although the pathophysiology of leptospirosis orchitis remains unclear, the present case suggests that this is a rarely noticed but likely manifestation of the disease in humans. Furthermore, it raises concern for the possibility of sexual transmission of leptospirosis, a disease which is seldom thought to be transmitted from person-to-person. Although the duration of persistence of leptospermia is unknown in either rams or humans, the patient was counseled to abstain from unprotected sexual intercourse for 6 months after infection in the event that viable spirochetes may be transmitted in sperm.

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