CASE REPORT

Scrub Typhus Complicated by Rare Human Pathogen *Sphingobacterium spiritivorum*

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Abstract

Sphingobacterium spiritivorum is a rare cause of human infections worldwide. After reviewing the literature, we could find only eight case reports to date. The majority of cases were of cellulitis and septicemia. Most of these patients were immunocompromised and the recovery rate was lesser. We present a case of a young female diagnosed with scrub typhus complicated by acute respiratory distress syndrome who developed septicemia and septic shock due to *S. spiritivorum*. She was managed with sensitive antibiotic levofloxacin, clinically improved, and discharged in satisfactory condition.

Keywords: Acute respiratory distress syndrome, Scrub typhus, *Sphingobacterium spiritivorum*.

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Introduction

Sphingobacterium spiritivorum is a rare cause of infection in humans. This bacterium has been associated with causing cellulitis,[1–4] bacteremia,[5,6] hypersensitivity pneumonitis,[7] sepsis, and infective endocarditis[8] in humans. Most reported cases were in immunocompromised patients or patients with multiple comorbidities. Here we present the first reported case of a young female with no comorbidities diagnosed as a case of scrub typhus with bacteremia attributing to *S. spiritivorum*, who clinically recovered well after appropriate antibiotic treatment.

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Case Description

A 37-year-old female with a single live baby and no comorbidities presented with complaints of persistent fever of 100°F for the last 5 days, increased frequency of stools for the last 3 days, and decreased urine output for the last 2 days. On examination, she was of average build, conscious, and oriented to time, place, and person. Her blood pressure was 80/50 mm Hg with a heart rate of 140/minutes (sinus tachycardia), and respiratory rate was 36/minutes with abdominal thoracic breathing. Laboratory investigations revealed—Hemoglobin—10 mg%, blood urea—84 mg%, serum creatinine—2.80 mg%, total leukocyte count (TLC)—14,300/mm^3 with 80% neutrophils, platelets—54,000/mm^3, sodium—125 mEq/L, potassium—3.1 mEq/L, PT-INR—1.18, SGOT—51 µ/L, SGPT—89 µ/L, total serum bilirubin—2.0 mg/dL, albumin 2.1 mg%. Fever workup was sent for tropical fevers and scrub typhus ELISA IgM reported positive. Her chest X-ray, 2D echocardiography, and ultrasonography of the abdomen were all normal. Fluid resuscitation and inotropic support were started with 0.9% normal saline and noradrenaline at 1.2 µg/kg/minute, respectively. The patient was put on noninvasive ventilatory support, and antibiotics meropenem, metronidazole, and doxycycline were added empirically. Right internal jugular venous cannulation was done. On day 2, IV fluids and noradrenaline were continued, as her shock was not responding vasopressin 0.04 µ/minute intravenously was added. Meanwhile, in 24 hour her blood, urine, and stool cultures reported no bacterial growth. She developed an oral mucosal bleed owing to worsening thrombocytopenia and it was managed with platelets transfusion. In view of oliguric acute kidney injury, she underwent one cycle of slow low efficient dialysis. On day 3, TLC got raised to 36,000/mm^3 with a neutrophilic predominance and she was started on teicoplanin. On day 4, the vasopressors' requirement decreased, and daily fluid adjustments were done. However later on day 4, her oxygen requirement increased because of worsening tachypnea and fatigue leading to mixed respiratory failure. Chest auscultation revealed coarse crepitations in bilateral lower zones. Chest X-ray now suggested pulmonary edema. Bedside echocardiography revealed decreased left ventricular ejection fraction (LVEF) of 40%. In view of severe acute respiratory distress syndrome (PaO_2/FiO_2 <100), she was kept on mechanical ventilation as per the lung-protective ventilation strategy. She was kept sedated and paralyzed. She received methylprednisolone 40 mg twice daily for 48 hours and then tapered off. In due course of time, on day 7 fractional oxygen requirement decreased to 40%. Vasopressors started tapering. Left ventricular ejection fraction (LVEF) improved to 60%, and chest X-ray, TLC, and serum creatinine showed an improving trend. A sedation vacation was given, and she regained consciousness but developed neuromuscular weakness in all four limbs. On day 8, fever again spiked to 103°F, TLC increased to 19,000/mm^3 with neutrophilic predominance, and vasopressors' requirement increased. Repeat blood culture and urine culture were sent. Antibiotics escalated to colistin and fluconazole.

Blood cultures after 2 days of incubation period reported the growth of gram-negative, non-motile, non-fermenting, oxidase-positive bacilli *S. spiritivorum*. Using the automated microdilution broth technique (vitek-2), antibiotic susceptibility testing was done. The results were reported as minimum inhibitory concentration (MIC): Cefoperazone/sulbactam ≤8 µg/mL, ciprofloxacin ≤1 µ/mL, levofloxacin ≤0.5 µ/mL, minocycline ≤1 µ/mL, trimethoprim/sulfamethoxazole ≤20 µ/mL. This bacterium was highly resistant to colistin, meropenem, imipenem, piperacillin/tazobactam, amikacin, gentamicin, cefepime, and ceftazidime. She was started on levofloxacin 500 mg intravenously once a day. On day 14 her fever subsided, vasopressors tapered off, and minimal oxygen requirement, and liberated from mechanical ventilation with no adverse event. Diagnostic workup was done for residual limb weakness and was diagnosed as critical illness-induced myopathy. Her clinical condition improved and was discharged in satisfactory condition on oral levofloxacin. Repeated blood cultures were done after 2 weeks of therapy and showed no growth of any organism. Her clinical events are summarized in Table 1.

Conclusion

We report the first human case of *S. spiritivorum* infection in a patient of scrub typhus. *Sphingobacterium* species mainly *S. multivorum* and *S. spiritivorum* are known to cause the majority of infections in humans. *S. spiritivorum* is a gram-negative rod, aerobic, non-fermenting, non-motile, non-sporing, oxidase-positive bacilli and was first described in humans by Holmes et al.[9] in 1982. After that only a few cases have been reported worldwide as described in Table 2. It is named *Sphingobacterium* because of the high content of sphingolipids in their cell walls. They are usually isolated from soil, water bodies, and plants and the isolation from human secretions is a rare event. In view of severe acute respiratory distress syndrome caused by scrub typhus, she received steroids which could have led to a pronounced immunocompromised state and supported the growth of bacteria in our patient. Interestingly she did not respond to empirical colistin and carbapenem initially as bacterial strains are inherently resistant to these antibiotics and the patient showed drastic recovery after receiving levofloxacin. We emphasize the use of repeated cultures and species identification among high-risk ICU patients as an utmost priority due to the intrinsic multidrug resistance behavior of this microorganism and increased risk of human infections.

Highlight

We present the first case of a young female with scrub typhus complicated by acute respiratory distress syndrome later on

Table 1: Patient timeline summarizing clinical events from presentation till discharge

Day 0	IV fluids, Inotropes, NIV, Antibiotics-meropenem, Doxycycline, Metronidazole
Day 1	Vasopressin started
Day 2	Platelets transfused for thrombocytopenia and dialysis given for acute kidney injury
Day 3	Antibiotic escalated to teicoplanin
Day 4	Inotropes reduced
Day 4	Features of myocarditis and ARDS, Steroids given
Day 7	Improvement in blood parameters, ARDS and myocarditis
Day 8	Fever spiked, Neutrophilic leukocytosis, Inotropes increased, Antibiotics escalated to colistin
Day 10	*S. spiritivorum* identified and levofloxacin started subsequently
Day 14	Clinical recovery, Inotropes tapered off, Weaning from ventilatory support
Day 18	Patient discharged in satisfactory condition on oral levofloxacin

ARDS, acute respiratory distress syndrome

Table 2: Summary of reported cases of *S. spiritivorum* in humans

Case	*Age (years)*	*Sex*	*Year*	*Comorbidity*	*Diagnosis*	*Source*	*Treatment*	*Outcome*
Marinella[1]	72	Male	2002	Parkinson	Cellulitis	Blood	NA	Recovery
Tronel[2]	84	Male	2003	Anemia	Cellulitis	Blood	Amoxicillin-clavulanate	Recovery
Anthony[3]	89	Male	2016	Parkinson	Cellulitis	Blood	Piperacillin tazobactam	Recovery
Arata[4]	80	Male	2017	COPD	Cellulitis	Blood	Levofloxacin	Recovery
Koh[5]	68	Female	2013	Leukemia	Septicemia	Blood	Ciprofloxacin	Death
Gupta[6]	80	Female	2016	End-stage renal disease	Septicemia	Blood	NA	Recovery
Kämpfer[7]	34	Female	2005	NS	Extrinsic allergic alveolitis	Water reservoir	NA	Recovery
Hugo[8]	61	Male	2016	Nephrotic syndrome	Infective endocarditis	Mitral valve	Piperacillin-tazobactam	Death
PC	37	Female	2021	NS	Septicemia	Blood	Levofloxacin	Recovery

PC, present case; NA, not available; NS, nothing significant

developing septicemia and septic shock due to the extremely rare human pathogen *S. spiritivorum*.

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