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CASE REPORT

First Case Reports of *Ignatzschineria* (*Schineria*) *indica* Associated with Myiasis

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We report three cases of infection due to the Gram-negative rod *Ignatzschineria* (*Schineria*) *indica* involving bacteremia and the urinary tract. Two cases were clearly associated with maggot infestation, and the third could conceivably have had unrecognized maggot infestation of the urinary tract. We believe these cases to be the first *I. indica* infections reported in association with maggot infestation and myiasis.

CASE REPORTS

Case 1 is a 64-year-old homeless male who presented to the emergency department at the University of Louisville Hospital with the chief complaint of a painful left foot. His pertinent medical history included a motor vehicle accident 2 months prior to this admission, at which time he sustained lacerations to the dorsal aspect of the first three digits of his left foot. Because of his social situation, he had been unable to treat his wounds or change the dressings since the accident. He complained of extreme pain in the foot, which was exacerbated with any movement or pressure. He reported no other symptoms or past medical history. On physical examination, his left foot was edematous and erythematous surrounding the bandages. Following removal of the dressings, the wound revealed malodorous lacerations located on the dorsum of the foot and along the border of digits 1, 2, and 3 which expressed serous drainage. Maggots were observed in the wound and between the digits. All pedal pulses were palpable. Vital signs and the remainder of the physical examination were unremarkable.

Laboratory studies revealed a normal white blood cell count (8,800/μl with 64.2% granulocytes), an elevated erythrocyte sedimentation rate (ESR [57 mm/h]) and elevated C-reactive protein (CRP) level (1.06 mg/dl). X ray of the left foot demonstrated mild dorsal soft tissue swelling with no acute fracture or dislocation. However, magnetic resonance imaging (MRI) of the foot showed a fracture of the third middle phalanx with adjacent soft tissue defect. The clinical impression was osteomyelitis, although not seen on imaging, and the patient was started on empirical ampicillin-sulbactam (3 g intravenous [i.v.] every 6 h [q6h]) and vancomycin (1.25 g i.v., q12h). His wounds were redressed wet to dry with Dakin's solution, and the necrotic tissue was debrided with removal of the maggots. Despite conservative treatment, the third digit was considered unsalvageable, and the patient was taken to surgery for amputation of the digit. Histopathology noted skin ulceration and prominent acute and chronic inflammation extending to the soft tissue margin. On the second day postadmission, two aerobic blood cultures were positive for nonhemolytic Gram-negative short plump rods. The isolate produced a "yellowish" pigment on blood agar. The oxidase and indole tests were, respectively, positive and negative. The organism was identified as *Alcaligenes faecalis* (97% probability) (RapidID NF Plus; Remel, Lenexa, KS). Attempts to perform susceptibility testing were unsuccessful due to the organism's not growing in the Microscan Gram-negative panels. The isolate was referred for 16S rRNA sequencing and was identified as *Ignatzschineria* (*Schineria*) *indica*, with an identity match of 99.44% (corresponding to a 4-bp mismatch).

The patient was started on cephalexin (500 mg *per os* [p.o.] 3 times a day [t.i.d.]), discharged on day 3, and lost to follow-up.

A living third-instar larva from case 1 was removed from the necrotic tissue associated with an open wound of the patient's foot and submitted for entomologic identification. The larva was placed in a sterile container to facilitate its growth to adulthood. Unfortunately, the larva expired before pupation and subsequently was fixed in 95% ethanol. The specimen was rehydrated and processed according to the method described by Cumming (1). The larva was identified as the blowfly, *Phaenicia sericata* (Meigen) (Diptera: Calliphoridae). This species and other members of the genus are among the most common flies infesting human wounds (2–4). These flies are cosmopolitan, relatively easy to maintain in the laboratory, effective at wound cleaning, and were commonly used to clean battlefield wounds in the Civil War and later; they commonly inhabit carrion and feces, are metallic green, and are frequently seen around meat, roadkill, and picnics (4–6).

Case 2 is a 67-year-old man with chronic alcoholism and extremely poor hygiene who was admitted to the Rapid City Regional Hospital with chronic nonhealing ulcers in the left heel with maggot infestation. A day prior to admission, he was seen at an-

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other hospital and was treated with clindamycin (900 mg) for suspected osteomyelitis. On admission, his physical examination and vital signs were unremarkable, with a temperature of 36.9°C, heart rate of 81, respiratory rate of 20, blood pressure of 121/71 mm Hg, and peripheral capillary oxygen saturation (SpO_2) of 96% at room air. The patient was alert, awake, and not in any acute distress but was mildly confused and disoriented. He was minimally communicative, with a blood alcohol level of 285 mg/dl. Other significant past medical history included a stab wound in the stomach and a recent (6 to 7 weeks prior) left supracondylar fracture which healed. On examination, dependent edema was noted on the left lower extremity, especially on the foot. The left foot was covered with multiple necrotic wounds, more extensively between the toes and heel. The skin between the first, second, and third toes extending down to the plantar surface was serrated, somewhat thickened, dark tan to brown, and appeared to be severely infected. The necrotic ulceration present on the lateral surface of the foot at the first metatarsal joint was deep and measured 3.5 by 3.0 cm. Sections through the first metatarsal joint exposed softened necrotic-appearing gray tan bone. Foul-smelling thick purulent discharge and significant maggot infestation were noted in those wounds. The largest ulceration was present on the lower lateral shin; it had an irregular geographic margin and measured 14.0 cm in length and up to 2.0 cm in diameter. Despite the unsalvageable condition of the foot, there was no substantial motor deficit to the extremities. On arrival, he was treated empirically with piperacillin-tazobactam and clindamycin. Radiographic images of the left foot indicated osteomyelitis. The patient underwent below the knee amputation of the left leg on the day of admission for management of clinical osteomyelitis and was treated with ciprofloxacin (500 mg p.o. 2 times a day [b.i.d.]) and vancomcyin (1 g i.v. q12h) postsurgery for 2 weeks.

Laboratory studies demonstrated a normal white blood cell count (6,900/μl), platelet count (320,000/μl), and glucose (107 mg/dl) and slightly decreased hemoglobin (10.9 g/dl) and hematocrit (33.9%). Blood cultures obtained at admission and during hospitalization remained sterile. However, blood cultures taken a day before admission at an outside hospital grew *Streptococcus pyogenes* and *Ignatzschineria indica*; the latter was initially identified by the Vitek 2 as an *Acinetobacter* species. Due to incompatible phenotypic characteristics, the isolate was submitted to a reference laboratory for identification by partial sequencing of the 16S rRNA gene, and the isolate was identified as *I. indica.* The target sequence showed 100% identity with the sequences corresponding to the 16S rRNA ribosomal gene of *I. indica.* The isolate was sensitive to amikacin (8 μg/ml), gentamicin (2 μg/ml), tobramycin (2 μg/ml), ceftazidime (8 μg/ml), cefepime (4 μg/ml), aztreonam (8 μg/ml), ciprofloxacin (≤0.12 μg/ml), levofloxacin (≤0.25 μg/ml), ticarcillin-clavulanate (≤4/2 μg/ml), and meropenem (≤0.25 μg/ml) and intermediate to piperacillin-tazobactam (34/4 μg/ml). All susceptibility interpretations were based on CLSI document M100-S23 (see Table 2B-5 in reference 7).

Case 3 is a 26-year-old male paraplegic. He was admitted to Parkland Memorial Hospital secondary to a gunshot wound that occurred 4 years prior to presentation. His accident resulted in a number of complications, including several unhealing decubitus ulcers for which surgical intervention for repair was denied by the patient on multiple occasions. He had resided in a number of long-term-care facilities and had poor management of his personal care and hygiene documented at multiple facilities. His medical history is notable for multiple past hospital admissions for urinary tract complaints, including recurrent urinary tract infections with positive urine cultures, ureteral and bladder stones, neurogenic bladder that required placement of bilaterial percutaneous nephrostomy tubes, and urethrocutaneous fistulas, which had allowed continuous leakage of urine through the decubiti. He was febrile on numerous admissions, accompanied by positive urine cultures consisting of *Escherichia coli*, *Proteus mirabilis*, vancomycin-resistant *Enterococcus faecalis*, *Pseudomonas aeruginosa*, *Providencia stuartii*, and on one occasion, an unidentifiable Gram-negative rod. During his most recent admission, a urine culture returned positive for a Gram-negative rod that was susceptible to aztreonam, ceftriaxone, cefepime, gentamicin, meropenem, trimethroprim-sulfamethoxazole, and tobramycin and had intermediate susceptibility to ciprofloxacin. Microscan (Siemens, Sacramento, CA) gave no identification, and the isolate was referred for 16S rRNA sequencing, which revealed *Ignatzschineria indica* with an identity match of 100%.

The genus *Ignatzschineria* (formerly known by the illegitimate name *Schineria*) belongs to the class *Gammaproteobacteria* and comprises three recognized species: *I. indica*, *I. larvae*, and *I. ureiclastica* (8, 9). All three species have been isolated from the obligate parasitic fly *Wohlfahrtia magnifica* (flesh fly; Diptera: Sacrophagidae), but we believe this is the first report of *I. indica* being associated with the blowfly. All three species are associated with myiasis, a disease of vertebrate animals caused by a variety of fly larvae. The members of this genus are aerobic, Gram-negative, non-spore-forming, nonhemolytic, nonmotile, rod-shaped bacteria. Colonies are small (0.05 to 0.2 mm), nonpigmented, entire, convex, and translucent, grow on blood agar, MacConkey agar, and *Salmonella-Shigella* (SS) agar, and are catalase and oxidase positive (9). Prior to our cases, the primary etiological agents reported in cases of myiasis were *Wohlfahrtiimonas chitiniclastica* and *I. larvae*, each associated with the parasitic fly *Wohlfahrtia magnifica. Wohlfahrtiimonas chitiniclastica* (originally identified by the Vitek system as *Brevundimonas diminuta* or *Oligella urethralis*), which also belongs to the class *Gammaproteobacteria* and is phylogenetically related to *I. larvae*, was recently reported as the cause of fulminant sepsis in a 70-year-old homeless man with a history of alcohol abuse and occlusive peripheral arteriopathy and a 60-year-old homeless woman with a history of alcoholism (10, 11). Two cases of bacteremia due to *I. larvae*, originally reported as *Schineria larvae*, were reported in a 76-year-old farmer with diabetes and myiasis of the leg, scrotum, and anus; and a 39-year-old homeless man with polyneuropathy related to alcohol abuse (12, 13).

Our cases highlight the importance of 16S rRNA amplification and sequencing to precisely identify microorganisms that are not in the databases of commercial automated devices or conventional biochemical identification systems that generate an identification probability of <90% (cases 1 and 2) or no identification (case 3). The observation that these organisms were recovered as the single agent in the respective blood cultures is suggestive of their invasive potential, which was enhanced by the fly larvae (maggots) serving as a vector when present in necrotic tissue and having direct access to the bloodstream.

Case 3 could not be definitively associated with active myiasis

due to the lack of recovery or observation of maggots at the time of admission; however, careful review of the patient's medical records and social history revealed dramatically similar risk factors (poor hygiene, substance abuse, and transient living conditions) to those described in the previously published literature and cases 1 and 2. Additionally, the cystoscopists noted difficulty in fully visualizing the bladder, and the patient refused additional exploration and debridement of the ducubitis ulcers; thus, maggots may have been present and not visualized. Given the scarcity of clinical cases reporting this bacterial genus and species and the overwhelming association with myiasis, it is likely that the patient in case 3 had urinary tract contamination due to myiasis at some time proximal to his admission. Given the unhealing nature of the decubitis ulcers, it is likely that this was the source for the organism to gain direct access to his urinary tract in addition to the presence of indwelling nephrostomy tubes with poor sanitary practices; myiasis would be the likely vehicle for this unusual finding in this scenario. In the absence of or failure to detect maggots, the recovery of *Ignatizschineria indica* in cultures may serve as an indicator of myiasis and promote further clinical investigation.

Ectoparasitism is common in homeless persons, especially body lice (*Pediculus humanus*) which are responsible for bloodstream infections, such as trench fever, epidemic typhus, and louse-borne relapsing. Our cases and those previously reported suggest that myiasis should be considered a form of ectoparasitism in homeless people and individuals with poor hygiene (10).

These cases further demonstrate the association of maggots and bacteremia. In cases of patients with poor personal hygiene and necrotic wounds displaying myiasis, blood cultures should be acquired to interrogate for possible *I. indica* infections. These cases also highlight the limitations of automated identification systems in accurately identifying organisms that are not included in their respective databases and support the value of submitting such isolates for definitive identification by 16S rRNA sequencing.

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