Case report

Ignatzschineria spp. bacteremia from maggot infestation

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A B S T R A C T

Ignatzschineria spp. bacteremia associated with maggot infestation is extremely rare in humans. There are only a few cases worldwide ever reported in the literature. We described a clinical case with a male patient who presented with maggot manifestation at his lower extremity, was found with bacteremia, and subsequently identified as Ignatzschineria spp by 16S RNA sequencing.

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Introduction

Ignatzschineria is a genus of aerobic, gram-negative, non-spore-forming, non-hemolytic rod-shaped bacteria that belongs to the class Gammaproteobacteria [1]. Three recognized species: l. indica, l. larvae, and l. ureiclasterica which are commonly recovered from the larvae of parasitic spotted flesh fly Wohlfahrtia magnifica found in Europe, Asia, and North Africa [2]. The Ignatzschineria spp. thrive in the digestive tract of the larvae with Providencia [2]. It is important to note that medical-grade maggots commonly used to promote debridement and wound healing are free of pathogenic bacteria as they are grown in sterile, cultured environments [3]. On the other hand, myiasis, the larval infestation of vertebrate animals and humans, occurs naturally in the environment and may cause uncontrolled infestation as well as bacterial infection [4].

Case study

A 63-year-old man with a history of hypertension and hyperlipidemia presented by EMS with a one-week history of malaise and left foot pain. The patient had his socks on for a week which he had trouble getting off as they seemed stuck to his skin. In the presentation, vital signs were significant with a temperature 101.8 °F, blood pressure 87/76, heart rate 113 beats/min, respiratory rate 21 breaths/min, and oxygen saturation >95 % on room air. Physical examination was unremarkable, except a 7 cm open wound on the anterior left ankle, deep down to the muscle layer, covered with numerous maggots (Fig. 1). The patient underwent irrigation and debridement with the wound vac placement. Blood culture on admission grew group C/G beta Streptococcus, Providencia stuartii, and finally, Ignatzschineria species were identified by 16S rRNA sequencing, PCR, and MALDI-TOF tests. Magnetic resonance imaging with the contrast of the left ankle revealed a large anterior deep ankle ulceration that transsects the tibialis anterior tendon, with no evidence of osteomyelitis (Fig. 2). The patient was initially started on vancomycin and cefepime, later changed to piperacillin/tazobactam for a total of 4 weeks of the regimen. Upon follow-up visit, the wound completely healed up (Fig. 3).

Discussion

A common cause of myiasis in animals, Ignatzschineria spp. rarely can cause human diseases, like myiasis and septicemia [2]. Wounds infected with maggots in the setting of poor hygiene were strongly associated with invasive Ignatzschineria spp. disease - with Ignatzschineria indica being the most common species identified. 16S rRNA amplification and sequencing, and mass spectrometry Matrix-Assisted Laser Desorption/Ionization-Time-of-Flight MALDI-TOF have been used in the identification of the bacteria, which are not typically included in standard microbiological identification systems [1]. However, some cases were described where MALDI-TOF was unsuccessful to identify the Ignatzschineria spp. [5,6].

There have been a few cases reported in Ignatzschineria spp. infection since 2014. Major risk factors in acquiring Ignatzschineria spp. infections include poverty, necrotic wounds, and poor personal hygiene. In the United States, cases of Ignatzschineria indica bacteremia have been described in patients who were disheveled, homeless, chronic alcoholics, local scraper, and bed-bound patients.
all had maggot-infested wounds in common [1,3,4,7]. Three cases of *Ignatzschineria indica* bacteremia in France were described in the setting of wounds infected with a large number of maggots [2]. A case of *Ignatzschineria indica* bacteremia was reported in the Netherlands in a patient with chronic alcoholism and maggot-infested wounds of the right foot [6]. Similarly, a case of *Ignatzschineria indica* septicemia associated with myiasis was described in Argentina and Spain, both patients were impoverished and chronic alcoholics, the former case led to the amputation of the patient’s left lower extremity [5,8]. A case of *Ignatzschineria indica* urinary tract infection was described in the United States in a paraplegic with chronic decubitus ulcers and urethrococutaneous fistulas with no observed maggot infestation. It is recommended that in the absence of maggots, culture isolation of *Ignatzschineria indica* or any organism commonly found in flies prompt investigation for occult myiasis [4]. Most of the cases were susceptible to

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**Fig. 1.** A large opened wound with significant maggot infestation.

**Fig. 2.** MRI showed a large anterior deep ankle ulceration, without osteomyelitis.

**Fig. 3.** Resolution of infection after the antimicrobial therapy.
beta-lactam antimicrobials, aminoglycosides and fluoroquinolones, however, there is currently no standardized antimicrobial susceptibility testing due to the rarity of this infection [4,8]. Subsequently, most of the cases were treated with beta-lactam antimicrobials based on sensitivity patterns, bacterial co-infections, and clinical response. (Table 1)

| Reference       | Geographic location | Patient background and presentation | Presence of maggots | Interventions                                                                                     | Outcome                  |
|-----------------|---------------------|-------------------------------------|---------------------|--------------------------------------------------------------------------------------------------|--------------------------|
| Barker et al., 2014 | United States       | Homeless with wounds on the left foot | Yes                 | - Blood                                                                                 | - Debridement            |
|                 |                     |                                     |                     | - Ignatzschineria indica (Confirmed using 16S rRNA sequencing)                             | - Empiric ampicillin-sulbactam and vancomycin de-escalated to cephalaxin |
|                 |                     |                                     |                     |                                                                                       | - Amputation of left, third toe |
|                 |                     |                                     |                     |                                                                                       | - Discharged              |
| Barker et al., 2014 | United States       | Chronic alcoholic with poor personal hygiene with nonhealing left heel ulcers | Yes                 | - Blood                                                                                 | - Empiric piperacillin-tazobactam and clindamycin switched to vancomycin and ciprofloxacine |
|                 |                     |                                     |                     | - Streptococcus pyogenes and Ignatzschineria indica (Confirmed using 16S rRNA sequencing) | - Below the knee amputation of left leg |
|                 |                     |                                     |                     |                                                                                       | - Amputated left leg      |
| Barker et al., 2014 | United States       | Paraplegic secondary to gunshot wound with chronic decubitus ulcers, uroseptic ulcer, fistulas | No                  | - Urine                                                                                 | - No data available       |
|                 |                     |                                     |                     | - Ignatzschineria indica (Confirmed using 16S rRNA sequencing)                             | - No data available       |
| Cipolla et al., 2017 | Argentina           | Homeless, chronic alcoholic with deep necrotic ulcer of left lower extremity | Yes                 | - Blood                                                                                 | - Debridement            |
|                 |                     |                                     |                     | - Ignatzschineria indica (Confirmed using 16S rRNA sequencing)                             | - Ciprofloxacin and clindamycin |
| Heddema et al., 2016 | Netherlands       | Alcoholic with wounds on his right foot found unconscious at home | Yes                 | - Blood                                                                                 | - ICU admission for non-invasive ventilation |
|                 |                     |                                     |                     | - Ignatzschineria spp. (Confirmed using 16S rRNA sequencing)                              | - Amoxicillin-clavulanic acid |
| Le Brun et al., 2015 | France             | Found unconscious in a forest, with maggots found around genital organs | Yes                 | - Blood                                                                                 | - ICU admission (had cardiopulmonary arrest) |
|                 |                     |                                     |                     | - Enterococcus faecalis, Enterobacter cloacae, Providencia stuartii, Corynebacterium spp., and Ignatzschineria spp. (Confirmed using 16S rRNA sequencing) | - Empiric ceftriaxone    |
| Lysaght et al., 2020 | United States     | Local “scrapper” with left foot and leg ulcers | Yes                 | - Blood                                                                                 | - Debridement            |
|                 |                     |                                     |                     | - Wohlfahrtimonas achitini-clastica, Ignatzschineria indica, and Providencia stuartii (data unavailable for method of confirmation) | - Empiric vancomycin, clindamycin and Piperacillin-tazobactam de-escalated to cefepime |
| Muse et al., 2017 | United States       | Bedbound for 6–8 months with decubitus ulcers, unresponsive and hypotensive | Yes                 | - Blood                                                                                 | - ICU admission for IV fluids, norepinephrine, mechanical ventilation |
|                 |                     |                                     |                     | - Streptococcus galolyticus, Streptococcus anginosus, and Ignatzschineria indica (data unavailable for method of confirmation) | - Debridement            |
| Rodriguez-Züniga et al., 2018 | Spain             | Impoverished, chronic alcoholic with wounds on right lower leg and left foot | Yes                 | - Blood                                                                                 | - Empiric vancomycin, cefepime and metronidazole de-escalated to levofloxacin |
|                 |                     |                                     |                     | - Ignatzschineria indica (Confirmed using MALDI TOF and 16S rRNA sequencing)              | - Amoxicillin-clavulanic acid |
| Snyder et al., 2020. | United States     | Disheveled with swollen and erythematous right lower extremity | Yes                 | - Blood                                                                                 | - Recovered              |
|                 |                     |                                     |                     | - Methicillin-resistant Staphylococcus aureus, Wohlfahrtimonas achitini-clastica and Ignatzschineria indica (Confirmed using MALDI TOF) | - Manual removal of maggots |
| Our case study: Do et al., 2021 | United States | Disheveled, erythematous, edema left lower extremity | Yes                 | - Blood                                                                                 | - Discharged recovered   |
|                 |                     |                                     |                     | - Ignatzschineria spp, group C/G beta Streptococcus, Providencia stuartii (Confirmed using MALDI TOF and 16S rRNA sequencing) | - Empiric vancomycin, cefepime, changed to Piperacillin-tazobactam |
|                 |                     |                                     |                     |                                                                                       | - Irrigation and debridement |
|                 |                     |                                     |                     |                                                                                       | - Discharged recovered   |

There is limited literature regarding the association of maggot species to bacteria causing invasive infection. Some studies raised the concern regarding the potential migration of Wohlfahrtimonas magnifica parasitic flies or if Ignatzschineria indica can potentially colonize other parasitic flies that cause myiasis [5,7]. A case of myiasis-induced sepsis from maggot infestation was described in the United
States where *Ignatzschineria indica* was isolated from blood culture and was believed to be caused by *Lucilia sericata* (green bottle fly) based on the appearance of the larvae [3]. Similarly, one of the *Ignatzschineria indica* bacteremia cases reported in the United States sent larvae for entomologic identification and was identified as the blowfly, *Phaenicia sericata* [1].

**Conclusion**

Maggot infestation is uncommon in the United States; therefore, diagnosis and management can be challenging. Cultures should be obtained for prompt *Ignatzschineria* spp. identification by 16S rRNA amplification and sequencing in patients at high risk for getting the infection, particularly if maggots present. Infection with *Ignatzschineria* is also considered with high-risk patients without obvious maggot infestation and should prompt investigation for occult myiasis.

**Declaration of Competing Interest**

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**Consent**

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request

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