



Case report

Ignatzschineria spp. bacteremia from maggot infestationSteven R. Do^{a,*}, Subhashis Mitra^a, Christopher Cantoria Garces^b, Farahnaz Anwar^b^a Division of Infectious Diseases, Department of Medicine, Michigan State University, College of Human Medicine, East Lansing, MI, USA^b Department of Medicine, Michigan State University, College of Human Medicine, East Lansing, MI, USA

ARTICLE INFO

Article history:

Received 1 May 2021

Received in revised form 4 May 2021

Accepted 4 May 2021

Keywords:

Ignatzschineria spp

Bacteremia

Maggot infestation

ABSTRACT

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 rRNA sequencing.

© 2021 The Author(s). Published by Elsevier Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

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: *I. indica*, *I. larvae*, and *I. ureiclastica* 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 transects 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

* Corresponding author.

E-mail address: dosteve1@msu.edu (S.R. Do).



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

- 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



Fig. 3. Resolution of infection after the antimicrobial therapy.

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 urethrocutaneous 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



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

Table 1
A list of all reported cases of *Ignatzschineria* spp. caused human infections in literature.

Reference	Geographic location	Patient background and presentation	Presence of maggots	Culture source, bacteria isolated and method of confirmation of <i>Ignatzschineria</i>	Interventions	Outcome
Barker et al., 2014	United States	Homeless with wounds on the left foot	Yes	- Blood - <i>Ignatzschineria indica</i> (Confirmed using 16S rRNA sequencing)	- Debridement - Empiric ampicillin-sulbactam and vancomycin de-escalated to cephalexin - Amputation of left, third toe	- Amputated left, third toe - Discharged
Barker et al., 2014	United States	Chronic alcoholic with poor personal hygiene with nonhealing left heel ulcers	Yes	- Blood - <i>Streptococcus pyogenes</i> and <i>Ignatzschineria indica</i> (Confirmed using 16S rRNA sequencing)	- Empiric piperacillin-tazobactam and clindamycin switched to vancomycin and ciprofloxacin - 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, urethrocutaneous fistulas	No	- Urine - <i>Ignatzschineria indica</i> (Confirmed using 16S rRNA sequencing)	- No data available	- No data available
Cipolla et al., 2017	Argentina	Homeless, chronic alcoholic with deep necrotic ulcer of left lower extremity	Yes	- Blood - <i>Ignatzschineria indica</i> (Confirmed using 16S rRNA sequencing)	- Debridement - Ciprofloxacin and clindamycin	- Left leg amputation - recovered
Heddema et al., 2016	Netherlands	Alcoholic with wounds on his right foot found unconscious at home	Yes	- Blood - <i>Ignatzschineria</i> spp. (Confirmed using 16S rRNA sequencing)	- ICU admission for non-invasive ventilation - Amoxicillin-clavulanic acid	- Discharged recovered
Le Brun et al., 2015	France	Found unconscious in a forest, with maggots found around genital organs	Yes	- Blood - <i>Enterococcus faecalis</i> , <i>Enterobacter cloacae</i> , <i>Providencia stuartii</i> , <i>Corynebacterium</i> spp., and <i>Ignatzschineria</i> spp. (Confirmed using 16S rRNA sequencing)	- ICU admission (had cardiorespiratory arrest) - Empiric ceftriaxone	- Found expired in his bed from no evident cause, despite initial clinical improvement 10 days after admission
Lysaght et al., 2020	United States	Local "scraper" with left foot and leg ulcers	Yes	- Blood - <i>Wohlfahrtiimonas chitiniclastica</i> , <i>Ignatzschineria indica</i> , and <i>Providencia stuartii</i> (data unavailable for method of confirmation)	- Debridement - Empiric vancomycin, clindamycin and piperacillin-tazobactam de-escalated to cefepime	- discharged recovered
Muse et al., 2017	United States	Bedbound for 6–8 months with decubitus ulcers, unresponsive and hypotensive	Yes	- Blood - <i>Streptococcus gallolyticus</i> , <i>Streptococcus anginosus</i> , and <i>Ignatzschineria indica</i> (data unavailable for method of confirmation)	- ICU admission for IV fluids, norepinephrine, mechanical ventilation - Debridement - Empiric vancomycin, cefepime and metronidazole de-escalated to levofloxacin	- Good clinical response - No other data available
Rodríguez-Zúñiga et al., 2018	Spain	Impoverished, chronic alcoholic with wounds on right lower leg and left foot	Yes	- Blood - <i>Ignatzschineria indica</i> (Confirmed using MALDI TOF and 16S rRNA sequencing)	- Amoxicillin-clavulanic acid	- Recovered
Snyder et al., 2020,	United States	Disheveled with swollen and erythematous right lower extremity	Yes	- Blood - <i>Methicillin-resistant Staphylococcus aureus</i> , <i>Wohlfahrtiimonas chitiniclastica</i> and <i>Ignatzschineria indica</i> (Confirmed using MALDI TOF)	- Manual removal of maggots - Empiric vancomycin and cefepime switched to daptomycin and ceftriaxone	- Discharged recovered
Our case study: Do et al., 2021	United States	Disheveled, erythematous, edema left lower extremity	Yes	- Blood - <i>Ignatzschineria</i> spp. group C/G beta <i>Streptococcus</i> , <i>Providencia stuartii</i> (Confirmed using MALDI TOF and 16S rRNA sequencing)	- Irrigation and debridement - Empiric vancomycin, cefepime, changed to piperacillin-tazobactam	- Discharged recovered

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)

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 *Wohlfahrtia 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

None

Funding

No sources of funding for this manuscript.

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

References

- [1] Barker HS, Snyder JW, Hicks AB, Yanoviak SP, Southern P, Dhakal BK, et al. First case reports of *Ignatzschineria* (*Schineria*) *indica* associated with myiasis. *J Clin Microbiol* 2014;52(12):4432–4, doi:<http://dx.doi.org/10.1128/JCM.02183-14>.
- [2] Le Brun C, Gombert M, Robert S, Mercier E, Lanotte P. Association of necrotizing wounds colonized by maggots with *ignatzschineria*-associated septicemia. *Emerg Infect Dis*. 2015;21(10):1881–3, doi:<http://dx.doi.org/10.3201/eid2110.150748>.
- [3] Lysaght TB, Wooster ME, Jenkins PC, Koniaris LG. Myiasis-induced sepsis: a rare case report of *Wohlfahrtiimonas chitiniclastica* and *Ignatzschineria indica* bacteremia in the continental United States. *Medicine (Baltimore)* 2018;97(52):e13627, doi:<http://dx.doi.org/10.1097/MD.00000000000013627>.
- [4] Snyder S, Singh P, Goldman J. Emerging pathogens: a case of *Wohlfahrtiimonas chitiniclastica* and *Ignatzschineria indica* bacteremia. *IDCases* 2020;19:e00723, doi:<http://dx.doi.org/10.1016/j.idcr.2020.e00723> Published 2020 Feb 15.
- [5] Cipolla L, Derooy L, Archuby D, Tarzia A, Govedic F, Prieto M. Sepsis secondary to complicated skin and soft tissue infection caused by *Ignatzschineria indica*. First case report in Latin America. *JMM Case Rep* 2018;5(6)e005151, doi:<http://dx.doi.org/10.1099/jmmcr.0.005151> Published 2018 Apr 17.
- [6] Heddema E, Janssen F, van Westreenen H. A case of *Ignatzschineria* bacteremia in an unconscious man from the Netherlands. *JMM Case Rep* 2016;3(3)e005043, doi:<http://dx.doi.org/10.1099/jmmcr.0.005043> Published 2016 Jun 25.
- [7] Muse H, Jenkins RL, Oliver MB, Kim S, Grantier RL, Malhotra BK, et al. A case of *Ignatzschineria indica* bacteremia following maggot colonization. *Case Rep Infect Dis* 2017;2017:3698124, doi:<http://dx.doi.org/10.1155/2017/3698124>.
- [8] Rodríguez-Zúñiga D, González-Galiano N, Leal-Negrado Á, Hidalgo-Pérez E. First case of sepsis by *Ignatzschineria* in Spain associated with myiasis. Description of a case and review of the literature. Primer caso de sepsis por *Ignatzschineria* en España asociada a miiasis. Descripción de un caso y revisión de la literatura. *Enferm Infecc Microbiol Clin* 2019;37(1):64–5, doi:<http://dx.doi.org/10.1016/j.eimc.2018.02.009>.