

### CASE REPORT

# **REVISED** Case Report: Shewanella algae, a rare cause of osteosynthesis-associated infection [version 2; peer review: 2 approved]

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**V2** First published: 13 Nov 2023, **12**:1465

https://doi.org/10.12688/f1000research.142096.1

Latest published: 02 Sep 2024, 12:1465

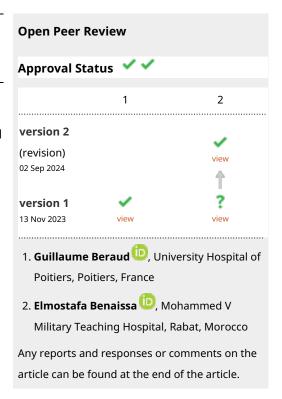
https://doi.org/10.12688/f1000research.142096.2

### **Abstract**

Shewanella is an emerging human pathogen. It mostly causes skin and soft tissue infections. Osteosynthesis-associated infection involving Shewanella are rare and in most cases are secondary to direct contamination following open fractures in aquatic environments. Here, we present a rare case of hematogenous osteosynthesisassociated infection involving Shewanella algae affecting an 18-yearold patient who was operated on for 12<sup>th</sup> thoracic vertebrae and 4th lumbar vertebrae fractures occurring in an aquatic environment. We performed surgical debridement with subsequent double course parenteral antibiotherapy that was then adapted to bacteria sensitivities for three weeks. After a follow-up of six months, the patient had no signs of recurrent infection. The presence of infected dermabrasions and the concordance between germs isolated in operative samples and in blood cultures presumes that the contamination was hematogenous.

# **Keywords**

Shewanella, Osteosynthesis, Infection, Osteosynthesis-associated infection, Hematogenous



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**Author roles: Masmoudi S**: Investigation, Writing – Original Draft Preparation; **Khlif MA**: Conceptualization, Supervision, Validation, Writing – Review & Editing; **Battikh H**: Resources, Validation; **Zribi M**: Resources, Validation; **Barsaoui M**: Conceptualization, Supervision; **Zitouna K**: Resources, Validation

**Competing interests:** No competing interests were disclosed.

**Grant information:** The author(s) declared that no grants were involved in supporting this work.

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How to cite this article: Masmoudi S, Khlif MA, Battikh H *et al.* Case Report: *Shewanella algae*, a rare cause of osteosynthesis-associated infection [version 2; peer review: 2 approved] F1000Research 2024, **12**:1465 https://doi.org/10.12688/f1000research.142096.2

First published: 13 Nov 2023, 12:1465 https://doi.org/10.12688/f1000research.142096.1

### **REVISED** Amendments from Version 1

In this second version of the article, we have emphasized in the presentation of the case that it was a closed fracture of the thoraco-lumbar spine associated with dermabrasions on both lower limbs, thus supporting our theory of hematogenous infection. We also specified the method used to identify this bacterial species.

Any further responses from the reviewers can be found at the end of the article

### **Background**

Shewanella is an aquatic Gram-negative bacillus and is widely found throughout the environment. The most commonly reported clinical presentation is skin and soft tissue infection, <sup>1-3</sup> often preceded by exposure to seawater. <sup>4</sup> Bacteremia is often found in premature neonates with congenital pneumonia, patients with infections of the soft tissues of the lower limbs and with underlying health issues such as chemical esophagitis, cholangitis and liver abscess. <sup>5</sup> All the cases of osteosynthesis-associated infection involving *Shewanella* reported in the literature occurred after a direct contamination following open fractures, most often in aquatic environments. <sup>6-9</sup> We report the first case of osteosynthesis-associated infection caused by *Shewanella algae* via haemathogenic route.

### Case presentation

An 18-year-old patient with no previous medical history of note was admitted to the intensive care unit after he fell into a well resulting in polytrauma. In addition to head and thoracic injuries, the whole body CT revealed a burst fracture of 12<sup>th</sup> thoracic vertebra with section of the spinal cord and complete paraplegia, burst fracture of 4<sup>th</sup> lumbar vertebra (Figure 1). Both fractures were closed. In cutaneous clinical examination we found multiple water-soiled dermabrasions in both legs.

He was operated on in the orthopaedic surgery department, and postero-lateral fusion was performed from the 10<sup>th</sup> thoracic vertebra to the 5<sup>th</sup> lumbar vertebra (Figure 2).

At the  $10^{th}$  post-operative day the patient presented fever (39.5°C), redness and swelling around the surgical wound with serous discharge (Figure 3). Dermabrasions in lower limbs were infected. The vital signs included blood pressure, 120/60 mm Hg (NR:  $\geq 90/60 \text{ mm Hg}$ ); respiration, 20 breaths per minute (NR: 12-18 breaths per minute); pulse, 95 beats per minute (NR: 60-100 beats per minute). Investigations showed a high white cell count ( $17.6 \times 10^9/L$ ) (NR:  $4.5-11 \times 10^9/L$ ) and a raised C-reactive protein (176 mg/L) (NR: <0.3 mg/L). Three blood cultures were performed.

The patient was reoperated on the 11<sup>th</sup> post-operative day. Intraoperatively, we found abundant pus with infected necrotic tissues that were then cleaned and debrided. We took five deep bacteriological samples. The operative wound



Figure 1. CT scan showing a burst fracture of 12<sup>th</sup> thoracic vertebra and the 4<sup>th</sup> lumbar vertebra.



Figure 2. Postoperative anteroposterior and profile radiography of the T10-L5 postero-lateral fusion.



Figure 3. A clinical photograph of the surgical wound showing inflammatory signs with serous discharge.

was closed on aspiratifs Redon drain. One of the blood cultures became positive, Gram staining performed from culture showed Gram-negative rods. They were identified as *Shewenella algae* by vitek 2. Intraoperative deep tissue specimens grew *Shewanella algae* and *Klebsiella pneumoniae*. *Shewanella algae* was resistant to amoxicillin, amoxicillin-clavulanic acid and levofloxacin, had intermediate susceptibility to trimethoprim-sulfamethoxazole and was sensitive to imipenem/cilastatin. *Klebsiella pneumoniae* was multi-resistant and was only sensitive to colistin. The patient had a double course of parenteral antibiotics (Imipenem/cilastatin at a dose of 500/500 mg/6 hours and colistin at a dose of 3 MUI/8 hours) for 25 days. The patient had minor adverse events such as epigastralgia and vomiting, which resolved with symptomatic treatment.

After three weeks of antibiotics, white cell count and C-reactive protein normalized. The surgical wound healed with no fistula. The patient was addressed to physical medicine and rehabilitation department. At eight months follow-up, the patient had no signs of recurrent infection.

#### Discussion

Shewanella has been regarded as an uncommon source of human infection. Despite being identified more than 70 years ago, our understanding of the bacterium's spread and the symptoms it causes comes primarily from a restricted set of individual case studies. Predominantly concentrated in tropical regions, the highest frequency of occurrences is noted within Southeast Asia, Southern Europe, and Africa. They naturally exist in various environments like water of all types, raw fish, oily food, and soils. Human infections involve Shewanella algae, putrefaciens, halitosis, and xiamenensis. However, the more offending species are Shewanella algae and putrefaciens accounting for more than 80% of cases. Shewanella infections can be serious leading to life-threatening conditions such as necrotizing fasciitis and septic shock. He route of infection is more likely cutaneous (wounds, leg ulcers, etc.), and, less frequently hepatobiliary or respiratory. Malignancy, hepatobiliary disease, diabetes, immunodepression, dysregulated iron metabolism and chronic infections of lower limb have been reported to be risk factors for developing a Shewanella infection. Although the patient received routine preoperative antibioprophylaxis based on 2 g of cefazolin and had no medical history, he developed infection.

In this case, *Klebsiella pneumoniae* was co-isolated in deep bacteriological samples. In fact, *Shewanella algae* are frequently identified in polymicrobial infections and the most common bacterial strains co-isolated are Enterobacteriaceae and marine flora bacteria.<sup>2</sup>

Cases of osteosynthesis-associated infection caused by *Shewanella* are rare. In our review of the literature, all cases were secondary to open fractures of lower limbs occurring in an aquatic environment. <sup>6,8,9,18</sup> To the best of our knowledge, this is the first case in which osteosynthesis implant contamination was secondary to bacteremia. *Shewanella algae* have a significant ability to haematogenous diffusion. Indeed, Vignier <sup>9</sup> and Yousfi <sup>19</sup> observed that bacteremia occurred in respectively 28% and 18% of the cases they studied. Mortality rates were respectively 13 and eight per cent. Bacteremia can lead to severe secondary infection including instances of epidural spinal abscess, purulent pericarditis, acute gastroenteritis accompanied by bloody diarrhea, and meningoencephalitis, as reported in various studies. <sup>20–23</sup> The concordance between germs isolated in operative samples and in blood cultures presumes that the contamination was haematogenous, probably originating from infected dermabrasions in both legs.

As in other cases of osteosynthesis-associated infection reported in the literature, we performed surgical debridement with subsequent double course parenteral antibiotherapy that was then adapted to bacteria sensitivities. Colistin was selected because it was the only effective antibiotic against *Klebsiella pneumonae*. Imipenem/cilastatin was the only antibiotic available in the hospital to which *Shewanella* was sensitive. Typically, *Shewanella* displays susceptibility to erythromycin, fluoroquinolones, chloramphenicol, third and fourth generation cephalosporins, aminoglycosides, carbapenems, and to some degree, trimethoprim-sulfamethoxazole and tetracyclines. However, it exhibits resistance against first and second generation cephalosporins, penicillin, and colistin.<sup>24</sup> An emergence of resistance has been documented towards imipenem and piperacillin/tazobactam, which can be attributed to the presence of the class D beta-lactamase enzyme.<sup>23</sup> Hopefully, our microbial stain was sensitive to imipenem/cilastatin.

Currently, there are no established guidelines for the management of shewanella infections. However, certain reports have indicated that addressing *Shewanella* infections may necessitate a proactive approach involving both surgical debridement and administration of appropriate antimicrobial agents. This particular case underscores the importance of recognizing *Shewanella algae* as a potential offending pathogen in osteosynthesis-associated infection coming within the framework of secondary hematogenous infection even in patients without significant underlying medical conditions.

### Consent

Written informed consent for publication of clinical details and clinical images was obtained from the patient.

### **Data availability**

All data underlying the results are available as part of the article and no additional source data are required.

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# **Open Peer Review**

# **Current Peer Review Status:**





# Version 2

Reviewer Report 04 September 2024

https://doi.org/10.5256/f1000research.170594.r319866

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### Elmostafa Benaissa 🗓



Mohammed V Military Teaching Hospital, Rabat, Morocco

The authors have responded to my comments. I thank them for their efforts in responding to these comments.

Competing Interests: No competing interests were disclosed.

**Reviewer Expertise:** medical microbiologymolecular biologyinfectious diseases

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

# **Version 1**

Reviewer Report 14 August 2024

https://doi.org/10.5256/f1000research.155595.r299831

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# Elmostafa Benaissa 🕛



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In this work by Masmoudi et al. a case of osteosynthesis-associated hematogenous infection involving the alga Shewanella is reported. The report is concise and addresses a case of infection by a relatively rare pathogen, but one for which there is a growing number of There have been only a few cases reported in the literature in recent years.

# **General comments:**

- Quality of presentation and structure of the manuscript: Satisfactory
- To what extent are the conclusions supported by the data: Satisfactory
- Do you have any concerns about possible image manipulation, plagiarism or any other unethical practice? : No
- If this manuscript involves human and/or animal work, have the subjects been treated ethically and have the authors followed appropriate guidelines? : Yes

# specific comments:

- authors should specify in their report the method used to identify this bacterial species (maldi-tof or classical method...)
- why did the authors speak of contamination via the blood-borne route, when the patient had open fractures exposed to aquatic, saprophytic and cutaneous flora? It may be that the osteosynthesis material was contaminated at the time of insertion and then infected, giving rise to bacteremia at the starting point of the osteosynthesis material. clarification is required in this case.

Is the background of the case's history and progression described in sufficient detail? Yes

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

**Partly** 

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?

Yes

Is the case presented with sufficient detail to be useful for other practitioners?  $\ensuremath{\text{Yes}}$ 

Competing Interests: No competing interests were disclosed.

**Reviewer Expertise:** medical microbiologymolecular biologyinfectious diseases

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Author Response 27 Aug 2024

### Sofiane Masmoudi

Dear Dr Elmostafa,

Thank you for reviewing our article. Your constructive comments made us aware of the lack of precision in certain aspects of our article. Indeed, it enabled us to give more details about microbiological techniques used to identify germs. Regarding your question about our theory of hemothogenic contamination, we have retained this theory given that the case involved a closed fracture of the thoracolumbar spine associated with water soiled dermabrations at a distance (on the both lower limbs) and the identification of shewanella algae on blood cultures. in this second version of the article, we have emphasized these elements in order to make the clinical case clearer for the reader.

**Competing Interests:** No competing interests were disclosed.

Reviewer Report 11 June 2024

https://doi.org/10.5256/f1000research.155595.r272741

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# Guillaume Beraud 🗓



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Masmoudi et al presents a case of Shewanella algae osteosynthesis-associated infection with the specificity that the pathogen was isolated in blood culture. They describe well the sequence that resulted in the infection, and their hypothesis is sound.

English is good, the manuscript is well written and easy to read.

I don't see any issue to be addressed.

Is the background of the case's history and progression described in sufficient detail?

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

Yes

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?

Yes

Is the case presented with sufficient detail to be useful for other practitioners?

Yes

**Competing Interests:** No competing interests were disclosed.

**Reviewer Expertise:** Infectious diseases

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

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