CASE REPORT

Wohlfahrtiimonas chitiniclastica-related soft-tissue infection and osteomyelitis: A rare case report

Mustafa Onur Karaca, M.D.,[1] Merve Gürler, M.D.,[2] Meltem Afacan, M.D.,[2] Mustafa Mert Terzi, M.D.,[1] Ebru Evren, M.D.,[2] Güle Çınar Aydın, M.D.,[3] Alper Tekeli, M.D.,[2] Mahmut Kalem, M.D.,[1] Zeynep Ceren Karahan, M.D.[2]

[1]Department of Orthopedics and Traumatology, Ankara University Faculty of Medicine, Ankara-*Türkiye*

[2]Department of Microbiology, Ankara University Faculty of Medicine, Ankara-*Türkiye*

[3]Department of Infectious Diseases and Clinical Microbiology, Ankara University Faculty of Medicine, Ankara-*Türkiye*

ABSTRACT

Wohlfahrtiimonas chitiniclastica is a rare pathogen that was first isolated from *Wohlfahrtia magnifica*, a parasitic fly. It is an uncommon, but an emerging human pathogen reported only in Europe and South America. Until today, it has been reported to be a zoonotic pathogen originating from different geographic locations. The present case, a patient suffering from osteomyelitis in Turkey, represents the first report of this pathogen in this country and so far no reports of related osteomyelitis associated with *W. chitiniclastica* is available. Clinical awareness of these emerging human pathogens is crucial for controlling infectious diseases.

Keywords: Osteomyelitis; soft tissue infection; *Wohlfahrtiimonas chitiniclastica*; zoonotic infection.

INTRODUCTION

Wohlfahrtiimonas is a genus of bacteria from the *Gammaproteobacteria* class and has two subspecies; *Wohlfahrtiimonas chitiniclastica* and *Wohlfahrtiimonas larvae*. The bacterium's name comes from a parasitic fly, *Wohlfahrtia magnifica*, which was first identified in 2008. These flies are located in France, Spain, Korea, Hungary, Egypt, and Turkey. Although *W. chitiniclastica* is a frequently reported infectious agent, it rarely causes human infections. While factors such as low economic income, poor hygiene, and alcoholism may predispose to bacterial infections, this bacterium has also been reported as an infection agent in farmers, elderlies, and people with paralysis. *W. chitiniclastica* cases were frequently detected in those living in relatively warm climates. The previous studies reported that the bacterium exhibits a spectrum of diseases ranging from simple wound infections to bacteremia resulting in septic shock and death in infected individuals.[1]

W. chitiniclastica is an aerobic, non-spore-forming, and non-motile Gram-negative bacillus that can grow between temperatures of 28–37°C.[2] It exhibits positive catalase and oxidase reactions with no urease activity, and no indole and hydrogen sulfide production. The presence of chitinase activity is defined as an important feature.[3]

Despite its known biochemical properties, the bacterium can be misidentified or omitted by conventional diagnostic methods. In recent years, with the increased use of proteomics-based bacterial identification methods such as matrix-assisted laser desorption/ionization time-of-flight mass spectrometry (MALDI-TOF MS) in medical microbiology laboratories, bacterial infections have been clarified more accurately. As a result, rare pathogens such as *W. chitiniclastica* can also be identified using MALDI-TOF MS (MALDI Biotyper® - RUO v.3.1; Bruker Daltonics GmbH and Co., Bremen, Germany).[2]

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Address for correspondence: Mustafa Onur Karaca, M.D.
Ankara Üniversitesi Tıp Fakültesi, Ortopedi ve Travmatoloji Anabilim Dalı, Ankara, Türkiye
Tel: +90 312 - 508 33 33 E-mail: karaca_onur@yahoo.com

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In this study, a polymicrobial wound infection and osteomyelitis accompanied with *W. chitiniclastica* on the right toe of a farmer dealing with livestock is reported.

CASE REPORT

A 57-year-old male patient was admitted to the orthopedics and traumatology outpatient clinic with a complaint of painless discharging wound on the right foot. During the physical examination, neither discharge nor warmth was detected at the wound site located posteromedially.

In the laboratory examination, the C-reactive protein level was 31.4 mg/L (reference: 0–5 mg/L), the white blood cell count was 6.46×10^9/dL (reference: $4.5–11\times10^9$/dL), and the procalcitonin level was 0.025 ng/mL (reference: <0.05 ng/mL).

The X-ray revealed a mild lytic bone lesion in the distal phalanx of the right toe with a blurred transition zone, which made us perform a magnetic resonance imaging (MRI) considering osteomyelitis (Fig. 1). A superficial ulceration in the medial plantar of the distal end of the right toe was observed in the MRI. There were accompanying inflammatory changes in the surrounding skin, subcutaneous tissues, and bone marrow edema compatible with osteomyelitis in the adjacent distal phalanx. The physical examination and radiological findings suggested a Meggitt–Wagner Grade 3 ulcerated wound in the patient (Fig. 1).

A swap sample and tissue biopsy samples from the wound were obtained to perform Gram staining and bacterial culture, which were carried out in the medical microbiology laboratory at our hospital. The patient was a 10-year ex-smoker with a 30-year smoking history, but had no history of alcohol use. He was diagnosed with rheumatoid arthritis 7 years ago, which was treated with sulfasalazine, hydroxychloroquine, and leflunomide, in addition to rituximab. There was no prominent feature in the family history.

In the microscopic examination of the Gram-stained preparation of the wound swab, no squamous epithelium cell or polymorphonuclear leukocyte was observed at ×10 magnification. At ×100 magnification, Gram-negative bacilli and Gram-positive cocci/coccobacilli morphology were observed. According to Gram-staining, the Q score was evaluated as 3.

For the isolation of fastidious and non-fastidious bacteria, the samples were cultured semi-quantitatively on sheep blood agar (SBA), eosin methylene blue agar (EMB), and BD™ chocolate agar (BD, Franklin Lakes, NJ, USA). The SBA and the EMB agar plates were incubated under aerobic conditions, and the chocolate agar was incubated in carbon dioxide environment at 37°C for 24–48 h.

After the incubation period, three different colony growths were detected. The non-pigmented colonies with a smooth center, rough edges, and shiny appearance on blood agar were defined as *W. chitiniclastica*, and the Gram-positive colonies were identified as *Streptococcus dysgalactia*e and *Arcanobacterium haemolyticum* with using the MALDI Biotyper® system (score ≥2).

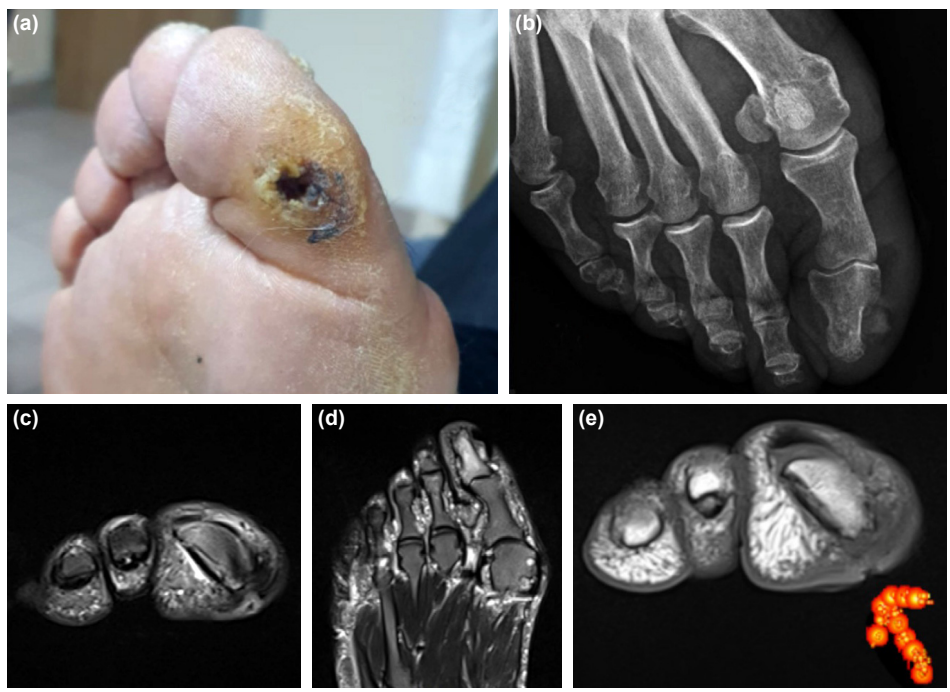

Figure 1. (a) A discharging wound on the posteromedial plantar aspect of the right first toe. **(b)** Mild lysis in the distal phalanx of the right toe. **(c)** A lytic lesion in the distal phalanx of the right toe. **(d)** Edema in the distal phalanx of the right toe. **(e)** Defective appearance on the skin (arrow).

Since *W. chitiniclastica* is a rare pathogen, the bacterium was also identified by 16S rRNA gene sequencing analysis. The 16S rRNA gene sequencing analysis result of *W. chitiniclastica* was also compatible with the MALDI-TOF MS result.

Antibiotic susceptibility testing was performed according to EUCAST 2020 guidelines and the results were reported as a MIC value with PK/PD comments for *W. chitiniclastica* and A. *haemolyticum*. *W. chitiniclastica* was found susceptible to all the tested antibiotics (Table 1).

The patient was treated with daily wound care and 10-day intravenous cefepime 2 g every 12 h. After discharge, the patient was advised to take 200 mg of cefpodoxime twice daily for 6 weeks. The patient had signed an informed consent before treatment.

DISCUSSION

W. chitiniclastica is a non-motile, non-spore-forming aerobic Gram-negative bacillus with catalases, and oxidases and can grow within a wide range of pH and temperature (pH: 5–10.5 and temperature: 28–37°C).[1,2,4]

As reported previously, particularly in Europe, Russia, and Africa, in myiasis cases, the microorganism was first isolated from the larvae of the *W. magnifica*.[5] The transmission of bacteria is thought to occur through the contact of adult *W. magnifica* fly or its larvae to mucosal surfaces and damaged skin.[5–8]

W. magnifica lives in limited areas around the world. The fact that the bacteria can be transmitted by *W. magnifica* larvae suggests that the prevalence of the infection may increase in those areas.[8] Therefore, it appears that almost all of the reported human infections, as in this case, were associated with open wounds and poor hygiene conditions.[7,8] Other risk factors included dealing with livestock, alcoholism, peripheral vascular disease, low socioeconomic status, and older age. Bueide et al.[8] stated that maggot therapy could also result in *W. chitiniclastica* infection. Therefore, the fact that our patient was a farmer dealing with livestock was considered an important risk factor for this infection.

Infections due to the aforementioned agent are often in the form of bacteremia, and cases reported as causative agents in wound infection are limited.[9] In reported cases, the agent often appears as a part of a polymicrobial infection.[1] In our case, it manifested itself as a member of polymicrobial infection in the form of *A. haemolyticum* and *S. dysgalactiae* at the wound site. As Kõljalg et al.[1] stated, the mechanism of the presence of *W. chitiniclastica* and other microorganisms in damaged tissues is not clearly known yet.

Besides the uncertainty of clinical appearance, the identification of the microorganism is still controversial. It has been reported that using traditional biochemical methods *W. chitiniclastica* can be misidentified as *Acinetobacter lwoffii*, *Comamonas testosterone*, or *Rhizobium radiobacter*.[1,2,4]

Actually, the identification of the microorganism can also be omitted as well. However, in recent years, the identification of the bacteria at the species level could be successfully done with MALDI-TOF MS and 16S rRNA gene sequencing. As a result, the widespread use of MALDI-TOF MS in routine diagnostic laboratories might provide us with a better knowledge to determine the effect of *W. chitiniclastica* in infectious diseases.

Due to the rarity of this infection, no antimicrobial treatment protocol has been standardized yet. *W. chitiniclastica* is sensi-

Table 1. Antimicrobial susceptibility testing for the three pathogens

Antibiotic	*Arcanobacterium haemolyticum*	*Streptococcus dysgalactiae*	*Wohlfahrtiimonas chitiniclastica*
Erythromycin		R[1]	
Imipenem (MIC), µg/ml			S[3]=0.25
Clindamycin		R	
Levofloxacin		I[2]	
Levofloxacin (MIC), µg/ml	S=0.38		S=0.064
Linezolid		R	
Moxifloxacin		S	
Penicillin		S	
Penicillin (MIC[5]), µg/ml	S=0.125		
Sefepim (MIC), µg/ml			S=0.125
Tetracycline		R	
Vankomisin (MIC), µg/ml	IE[4]		

[1]R: resistant; [2]I: Intermediate; [3]S: susceptible; [4]IE: insufficient evidence; [5]MIC: Minimum inhibitory concentration.

tive to most classes of antimicrobials, including beta-lactams, aminoglycosides, fluoroquinolones, and tetracyclines.[6,9] Usually, a combination of antibiotics from beta-lactam and aminoglycoside or quinolone groups is used to treat this unique microorganism.[8,9]

The current treatment of *W. chitiniclastica* infection includes the complete removal of larvae with sequential debridement in addition to antimicrobials. In some cases, where myiasis is severe, debridement may be extensive, and amputation may be required.[6,7] In case the blood cultures grow *W. chitiniclastica*, antibiotics should be administered for at least 1 week intravenously, and later orally.[8]

Conclusion

Although *W. chitiniclastica* has been frequently reported to cause bacteremia, it should also be considered as a predisposing factor in soft-tissue infections. To the best of our knowledge, this soft-tissue infection and osteomyelitis case are also the first report from Turkey. The correct identification of the bacterium will contribute to the literature and will shed light to the cases with various infectious diseases.

Informed Consent: Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

Peer-review: Internally peer-reviewed.

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Conflict of Interest: None declared.

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OLGU SUNUMU - *ÖZ*

Wohlfahrtiimonas chinisclastica ilişkili yumuşak doku enfeksiyonu ve osteomiyelit: Nadir bir olgu sunumu

Dr. Mustafa Onur Karaca,[1] Dr. Merve Gürler,[2] Dr. Meltem Afacan,[2] Dr. Mustafa Mert Terzi,[1] Dr. Ebru Evren,[2] Dr. Güle Çınar Aydın,[3] Dr. Alper Tekeli,[2] Dr. Mahmut Kalem,[1] Dr. Zeynep Ceren Karahan[2]

[1]Ankara Üniversitesi Tıp Fakültesi, Ortopedi ve Travmatoloji Anabilim Dalı, Ankara
[2]Ankara Üniversitesi Tıp Fakültesi, Tıbbi Mikrobiyoloji Anabilim Dalı, Ankara
[3]Ankara Üniversitesi Tıp Fakültesi, Enfeksiyon Hastalıkları ve Klinik Mikrobiyoloji Anabilim Dalı, Ankara

Wohlfahrtiimonas chitiniclastica, ilk olarak parazitik bir sinek olan *Wohlfahrtia magnifica*'dan izole edilen nadir bir patojendir. Nadir görülen ancak yeni ortaya çıkan bir insan patojenidir ve yalnızca Avrupa ve Güney Amerika'da rapor edilmiştir. Bugüne kadar farklı coğrafi konumlardan kaynaklanan zoonotik bir patojen olduğu bildirilmiştir. Türkiye'de osteomiyelitten muzdarip bir hasta olan mevcut olgu, bu ülkedeki bu patojenin ilk olgusudur ve şimdiye kadar *W. chitiniclastica* ile ilişkili osteomiyelit ile ilgili herhangi bir rapor mevcut değildir. Ortaya çıkan bu insan patojenlerine ilişkin klinik farkındalık, bulaşıcı hastalıkları kontrol etmek için oldukça önemlidir.

Anahtar sözcükler: Osteomiyelit; *Wohlfahrtiimonas chinisclastica*; yumuşak doku enfeksiyonu; zoonotik enfeksiyon.

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