# A Case of Wohlfahrtiimonas Chitiniclastica Contributing to Polymicrobial Osteomyelitis in the United States

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## Abstract

*Wohlfahrtiimonas chitiniclastica* is a rare emerging bacterium first isolated from the larva of *Wohlfahrtia magnifica* that has most commonly been associated with myiasis. This emerging human pathogen has spread to several geographical locations, with more cases being reported in the United States. Although rare, isolated cases of *W. chitiniclastica* causing bacteremia, cellulitis, and osteomyelitis have been reported. We present a case of *W. chitiniclastica* as a component of polymicrobial osteomyelitis in a 37-year-old male in Oklahoma, US, of which the patient was successfully treated with daptomycin and ertapenem after debridement.

## Introduction

First isolated from the larva of *Wohlfahrtia magnifica*, *Wohlfahrtiimonas chitiniclastica* is a rare gram-negative bacterium that is often found in areas of poor sanitation, particularly within developing countries. [15] Originally, reports of myiasis from parasitic flies were localized to the tropical regions.[7] However, other case reports have shown an increase in infections to other more temperate regions. While not yet widely studied, *W. chitiniclastica* is phylogenetically similar to *Ignatzchineria* larvae, which is known to cause myiasis. [10] *W. chitiniclastica* has been isolated from the gut of the house fly *Musca domestica* [13]. W. chitiniclastica have been known to cause myiasis within animals such as sheep and cattle. [4,15] *W. chitiniclastica* are not limited to the *Wohlfahrtia magnifica* flies, as it has been also found within *Chrysoma megacephala*, *Lucilia sericata* (green bottle fly), and *Hermetia illucens* flies. [4,12]

The first case of *W. chitiniclastica* reported in humans was recorded in France in May 2006. In this case, a homeless woman with alcoholism was found to have scalp excoriations with lice and larval infestation, and it was believed that her bacteremia originated from the scalp maggots. [10] In Japan, a man with a chronic left shoulder wound due to squamous cell carcinoma was found to have *W. chitiniclastica* bacteremia in conjunction with multiple other organisms after initially having negative blood cultures for the pathogen on admission.[6] Recently, a case of *W. chitiniclastica* was reported to have caused cellulitis and deep ulcer formation of the right foot which ultimately progressed to osteomyelitis in Kerala, India.[13] The majority of patients who have been infected with the bacterium have had poor hygiene, alcohol dependence, and chronic wounds.

In the United States, the first documented case of human infection from *W. chitiniclastica* was right leg swelling with draining ulcers in 2015.[5] The patient's occupation put him at a higher risk of being in contact with insects, and his morbid obesity made properly cleaning his wounds difficult.[5] Since the first case of *W. chitiniclastica* was reported, there have been additional reports around the United States. In the continental US, a 37-year-old male with ulcers on his left lower extremity presented with polymicrobial myiasis including *W. chitiniclastica.*[8] In Hawaii, two elderly patients had *W. chitiniclastica* sepsis secondary to skin and soft tissue infections, with one case being fatal. Both of the patients were living under poor hygienic conditions. *W. chitiniclastica* was isolated from blood cultures and maggots were noticed in the wound in one of two patients; however, a coexisting *E. coli* infection could have contributed to the death of the patient. [9]

There are various methods used in identifying the *W. chitiniclastica* bacterium. The matrix-assisted laser desorption ionization-time of flight mass spectrometry (MALDI-TOF MS) has been used for identification, and 16S rRNA gene sequencing has been utilized to confirm the sequencing to determine the presence of *W. chitiniclastica* within multiple reports involving the bacterium. [1,2,14] After diagnosis, treatment of *W. chitiniclastica* involves the removal of the larvae and usage of antimicrobials. *W. chitiniclastica* has been shown to be susceptible to most classes of antibiotics with beta-lactams, aminoglycosides, carbapenems, and fluoroquinolones being the most commonly used. [3,11] Only one reported case has shown the bacterium to have mild resistance to tetracyclines, [12] and there has been some intrinsic resistance to fosfomycin not yet studied. [11]

**Case Report**

We present a 37-year-old Oklahoma male with poorly controlled diabetes mellitus and chronic foot wounds with frequent readmissions secondary to wound infections. Per medical chart review of all admissions, the patient was never found to have mention of poor hygiene, farm animal exposure, or a distinctive living situation. He was first seen at Oklahoma State University Medical Center (OSUMC) in September 2019 for a chief complaint of right foot erythema and edema that began approximately two weeks prior that slowly spread superiorly. The patient had been seeing a podiatrist for a noted chronic diabetic foot ulcer, but he had not undergone any intervention. During this presentation, the patient was septic secondary to a right foot wound which was draining malodorous purulent material with tracking to the deep tissues. A CT scan of the right foot at that time showed subcutaneous air between the first and second toes, and the patient was placed on vancomycin, piperacillin/tazobactam, and clindamycin for empiric antibiotic coverage. Additionally, General Surgery was consulted for operative management. During this initial hospitalization, the patient underwent two surgical debridements, wound vacuum placement, and PICC placement for prolonged antibiotic therapy with ciprofloxacin. He was then transferred back to the hospital from which he initially was directly admitted for wound management and completion of antibiotics. However, the patient did return to OSUMC after approximately 14 days for a repeat MRI of the right foot, which revealed evolving osteomyelitis of the second metatarsal and second proximal phalanx and extensive osteomyelitis in the right great toe. Despite worsening imaging findings, the patient at that time endorsed “feeling better.” Finalized cultures from the wound demonstrated *Streptococcus agalactiae*, ampicillin-sensitive *Enterococcus*, unspeciated diphtheroids, and *Bacteroides fragilis*. The patient was then placed on piperacillin/tazobactam for 6 weeks and returned to his local hospital for ongoing treatment.

One year later, the patient again presented to OSUMC as a direct admit from an outlying hospital in southeastern Oklahoma for a suspected diagnosis of osteomyelitis given the presence of foot pain, fever, and chills. Patient reported noticing significantly more swelling of his right foot over a span of several months and had not been following up with any wound care. He presented in septic shock and was found to have beta Streptococcus present on blood cultures. X-ray of the right foot demonstrated osteomyelitis again of the second right metatarsal and right second phalanx. Patient was volume resuscitated and started on linezolid and piperacillin/tazobactam. Following admission, General Surgery was consulted and performed a debridement followed by amputation of the disarticulated right 2nd phalanx. Upon conclusion of this procedure, a wound

vacuum was placed. Repeat blood cultures were negative and wound cultures did show *Streptococcus dysgalactiae* as well as *Parvimonas micra*. A bone culture was also obtained and once finalized, showed growth of *Streptococcus dysgalactiae*, diphtheroids, *Parvimonas micra*, and a non-fermenting gram-negative rod. Further testing of the non-fermenting gram-negative rod by matrix-assisted laser desorption and ionization (MALDI) revealed colonies of *W. chitinclastica*. The patient was given ertapenem for six weeks and transferred to a local hospital for wound care and completion of antibiotics. He was readmitted the following day after an MRI demonstrated osteomyelitis despite amputation with the presence of sero-sanguineous drainage from the surgical site. General Surgery deemed no further surgical intervention was necessary. Infectious Diseases was consulted with recommendations to broaden treatment with the addition of daptomycin for coverage of diphtheroids. The patient was discharged on both ertapenem and daptomycin without current readmission to date.

**Discussion**

Currently, there is limited literature on *W. chitiniclastica* in patients with cellulitis and osteomyelitis. However, the pathogenicity of *W. chitiniclastica* continues to be researched as more cases involving this bacterium are published. First discovered as phylogenetically similar to *Ignatzschineria,* [7] it is classified as a non-motile, strictly anaerobic, Gram negative rod that exhibits both catalase and oxidase positive activity. [3,11] Because of its similarity to *Ignatzschineria* in both strong chitinase activity and preferred environment, it has potential to cause co-infection such as bacteremia. [8] It has been shown that transmission of *W. chitiniclastica* occurs when fly larvae are deposited into damaged skin and soft tissues, such as open wounds .[12] Based on previous case reports, it seems that common risk factors for *W. chitiniclastica* include poor hygiene, alcohol abuse, peripheral vascular disease, and chronic open wounds that are often untreated. [5,6] While no maggots were noted on physical examination during any of his hospitalizations, the patient in this case report had a chronic open wound for several months with poor wound care. The patient did have concern for bilateral vessel hardening of the lower extremities seen on ankle-brachial indices, with concern for ischemia of the right foot on repeated toe-brachial indices.

The patient in this case was successfully treated with ertapenem and daptomycin. In previous studies, *W. chitiniclastica* has generally been shown to be pan-sensitive to several classes of antibiotics, with only one documented case of mild resistance to tetracyclines. [4,9,12] It has exhibited some mild intrinsic resistance to Fosfomycin which has yet to be seen or tested with treatment of human infections.[11] The most common classes of antibiotics used to treat *W. chitiniclastica* have included beta-lactams, carbapenems, aminoglycosides, and fluoroquinolones,[4] of which a carbapenem was used in this particular case of osteomyelitis. It can be presumed that minimal resistance is secondary to the youth of the infection in humans and minimal exposure to antibiotics thus far.

Of the patients presented in other case reports, it appears that mortality is related to the presenting condition of the patient (i.e. fulminant sepsis) rather than *W. chitiniclastica* itself [11]. In one of two documented cases of *W. chitiniclastica* in Hawaii, US, a 72-year-old male who was found unattended after three days with an umbilical maggot-infested wound died on day 2 of

hospitalization [9]. Another patient with *W. chitiniclastica* bacteremia in a 41-year-old female was found to have Candida albicans fungemia after death.[3] A case of *W. chitiniclastica* bacteremia in a 70-year-old male resulted in fulminant septic shock in which the patient required intubation and vasopressors in the ICU.[1] In the presented case above of osteomyelitis, source control was obtained via debridement and amputation leading to overall clinical improvement of the patient. *W. chitiniclastica* has been reported in both monomicrobial and polymicrobial infections. Case reports have shown *W. chitiniclastica* has been isolated with bacterium such as *Escherichia coli*, *M. morganii*, *Proteus mirabilis*, *Providencia rettgeri*, and *Staphylococcus aureus.* [4] Mortality also appears to not be affected by presence of other organisms, as patient deaths were reported in monomicrobial *W. chitiniclastica* bacteremia as well as in polymicrobial wound infections. [1,9] With polymicrobial infections, such as the right foot osteomyelitis studied here, blame cannot be directly assigned to *W. chitiniclastica* .[2,5]

It is important to note that proper identification of *W. chitiniclastica* is necessary for its diagnosis. Initial microbiological methods can often lead to misdiagnosis due to it having characteristics similar to other organisms. It has previously been miscultured as *Acinetobacter lwoffii* [5] and Comamonas using the VITEK-2 system. [7, 12] *W. chitiniclastica* has been successfully and properly identified by MALDI in previous case reports, as was done at OSUMC. In this particular patient with recurrent osteomyelitis, he did not have growth of *W. chitiniclastica* initially. Similarly, a case of a 75-year-old male with a necrotic, maggot-infested shoulder wound in Japan reported growth of *W. chitiniclastica* not until the twentieth day of hospitalization. In this case, it was suspected that the bacterium entered the patient during rather than prior to his hospitalization. [6] Since *W. chitiniclastica* may not always be present in initial cultures, it is important to keep it on the differential diagnosis in patients with concerning risk factors.

**Conclusion**

In conclusion, we have reported a case of a rare emerging bacterium, *W. chitiniclastica* that was associated with polymicrobial osteomyelitis. While initially seen in warmer climates, it is now being recognized as a potential pathogen for bacteremia, skin/soft tissue infections, and osteomyelitis. It is important to raise clinical awareness of the risk factors most commonly associated with *W. chitiniclastica* and encourage patients of the importance of local wound care to prevent further infection and complications.

**Disclaimers**

The opinions expressed by authors contributing to this journal do not necessarily reflect the opinions of the Oklahoma State University or the institution with which the authors are affiliated.

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