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CASE REPORT

Kerstersia gyiorum-Caused Chronic Osteomyelitis in a Male Patient with Cerebral Infarction: A Case Report and Literature Review

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Abstract: *Kerstersia gyiorum* (*K. gyiorum*) had been reported as a rare cause of human infections. In this study, we isolated an uncommon strain of *K. gyiorum* from swab specimens of a male patient with cerebral infarction. The identification was performed using VITEK MS in the RUO (Research Use Only) mode and 16s rRNA gene sequencing. We hereby report a case of chronic osteomyelitis caused by *K. gyiorum* in a patient with cerebral infarction and left-sided limb hemiplegia in China.

Keywords: *Kerstersia gyiorum*, chronic osteomyelitis, VITEK MS

Introduction

Kerstersia gyiorum, first described by Coenye et al in 2003, is a Gram-negative bacillus with a small size (1–2 μm), flat morphology, positive catalase and negative oxidase activities; however, the activities of urease and β-galactosidase remain under investigation.[1] It grows at temperatures between 28°C and 42°C, and its genomic G+C content ranges from 61.5% to 62.9 mol%.[1] The bacterium has been isolated from clinical specimens such as ear swabs, wound secretions, blood, sputum, and urine.[2–6] Nevertheless, only a limited number of human infections have been reported to date, indicating that it is rare in humans.

The genus *Kerstersia* belongs to the family *Alcaligenaceae* and is phylogenetically related to the genera *Alcaligenes, Achromobacter, Bordetella*, and *Pigmentiphaga*.[1] Based on 16S rDNA sequence analysis, *Kerstersia* shows similarity to *Bordetella* species (93.5–96.4%), *Achromobacter* species (91.6–95.1%), *Alcaligenes* species (92.7–94.8%), and *P. kullae* (92.9–93.5%).[1] Phenotypically, *K. gyiorum* resembles *A. faecalis*.[1] Although biochemical identification alone is insufficient to differentiate *Kerstersia* from other *Alcaligenaceae*, whole-cell fatty acid analysis can distinguish it by the absence of 12:0 2-OH and the presence of high relative amounts of 18:1w7c.[1]

In 2012, Vandamme et al reported a second species, *Kerstersia similis*, isolated from a neck abscess.[7] *K. gyiorum* and *K. similis* cannot be reliably distinguished by biochemical methods, as they share 99.3% similarity in 16S rRNA gene sequences. However, *gyrB* gene sequence similarity ranges from 97.2% to 98%, and (GTG) 5-primed PCR fingerprinting allows clear differentiation between the two species.[7] The species name "Gyiorum" is derived from the Greek word for "limb", while "smilis" (from Latin) means "similar", reflecting the biochemical resemblance between the two species.[1,7]

Chronic osteomyelitis is a refractory bone infection that typically requires combined medical and surgical treatment. It is prone to recurrence even after successful treatment and may reactivate during quiescent phases. Currently, there is no generally accepted classification system for chronic osteomyelitis, and no clear time threshold defines when a bone infection becomes chronic. Therefore, chronic osteomyelitis remains a challenging condition for clinicians.[8] Here, we report a case of chronic osteomyelitis caused by *K. gyiorum* in a Chinese patient with cerebral infarction and left-sided hemiplegia.

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Case Presentation

A 64-year-old male patient with chronic osteomyelitis was admitted to our hospital due to persistent discharge from the right lower leg. Physical examination revealed mild swelling of the right lower limb with pigmentation of the surrounding skin, as well as two ulcerations on the anterior tibia measuring approximately 1 cm and 0.5 cm in diameter, respectively (Figure 1A). MRI findings were consistent with chronic osteomyelitis (Figure 1B), showing internal purulent-like tissue accompanied by fluid drainage. The patient had sustained a right tibial shaft fracture more than 10 years ago, which was initially treated with plate internal fixation. The hardware was subsequently removed due to purulent discharge. During this period, he received empirical antibiotic therapy at other hospitals, which temporarily alleviated symptoms. However, over the past two months, his condition worsened with increased ulceration and purulent secretion, and antibiotic treatment at another hospital proved ineffective. The patient has a history of hypertension for over 10 years and has experienced purulent and fluid discharge from the right lower leg for more than a decade. More than one year ago, he suffered from a cerebral infarction resulting in left-sided hemiplegia.

Upon admission, inflammatory markers were elevated (Table 1), including interleukin-10 (IL-10), interleukin-6 (IL-6), high sensitivity C-reactive protein (hs-CRP), procalcitonin (PCT), and erythrocyte sedimentation rate (ESR).

Following debridement, two swab specimens were collected from the sinus tract and sent for microbiological analysis. After 24 h of incubation at 35 °C in 5% CO_2, significant pure bacterial growth was observed on blood agar and MacConkey agar (Figure 2).Colonies on blood agar (Figure 2A) appeared flat, grayish-white, and dry, while on MacConkey agar (Figure 2B) were flat, light pink, and dry. Initial identification using VITEK MS (bioMerieux, France) in IVD (In Vitro Diagnostic) mode was unsuccessful, although a characteristic spectrum was detected. Subsequent analysis using VITEK MS in RUO (Research Use Only) mode identified the isolate as *K. gyiorum* (Figure 3). This result was confirmed by matrix-assisted laser desorption/ionization-time of flight mass spectrometry (MALDI-TOF MS; Bruker Daltonics, Bremen, Germany) using a MALDI Biotyper system. Identification attempts with the VITEK 2XL (bioMerieux, France) were also unsuccessful, though eight biochemical reactions (ProA, ILATk, SUCT, IMLTa, TyrA, CIT, CMT, and ILATa) yielded positive results.

Given the rarity of *K. gyiorum* in human infections, 16S rRNA gene sequencing was performed for confirmation. Bacterial DNA was extracted using the TSINGKE Plant DNA Extraction Kit (Universal) (Tsingke Biotechnology Co., Ltd., Beijing). Amplification was carried out with primers 27F (GAGAGTTTGATCCTGGCTCAG) and 1492R

Figure 1 Ulcerations at time of admission (**A**), MRI image of chronic osteomyelitis (**B**).

Table 1 Inflammatory Indicators of the Patient at Time of Admission

Indicator	Result
White blood cell (WBC)	6.79×10^9/L
Interleukin-10 (IL-10)	5.92pg/mL
Interleukin-6 (IL-6)	5.36pg/mL
High sensitivity C - reactive protein (hs-CRP)	5.5mg/L
PCT	0.06pg/mL
Erythrocyte Sedimentation Rate (ESR)	44mm

(TACGGCTACCTTGTTACGAC) under the following conditions: pre-denaturation at 98°C for 3 min; 39 cycles of denaturation at 98°C for 10s, annealing at 55°C for 15s, and extension at 72°C for 15s;and a final extension at 72°C for 5 min. Sanger sequencing results showed 99.93% identity with *K. gyiorum*strain WCHKG1 in the NCBI GeneBank Database (https://blast.ncbi.nlm.nih.gov/Blast.cgi).

Antimicrobial susceptibility testing was carried out using the BD Phoenix™ M50 (Becton, Dickinson and Company, America). MIC values are summarized in Table 2. Based on identification and susceptibility results, the patient was treated with Ceftriaxone (2g, once daily). After ten days of treatment, purulent discharge decreased, ulcer sizes reduced, and granulation tissue formation was observed. Surgical intervention was recommended but was declined by the patient. At discharge, his condition had improved significantly, although complete healing of the sinus tract remained challenging due to chronic osteomyelitic changes. Telephone follow-up indicated that the patient remained in stable condition with only occasional minor pus discharge.

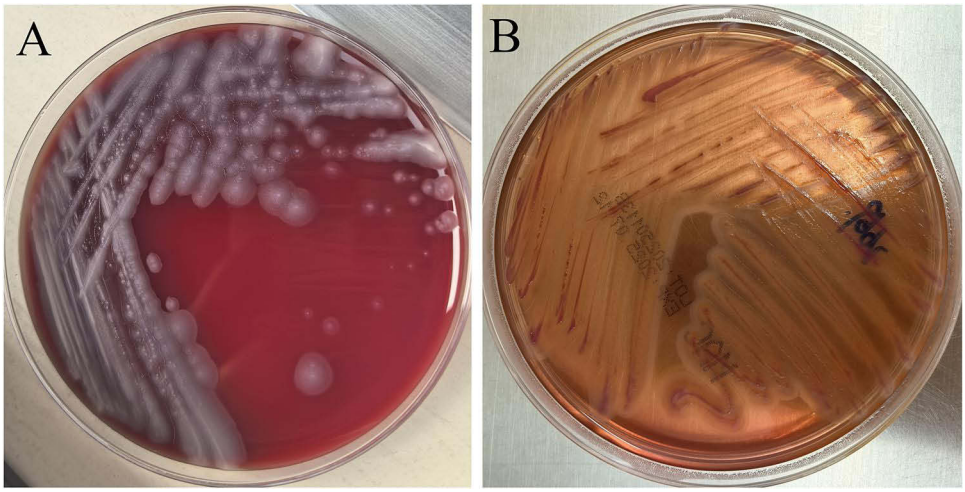

Figure 2 Colony morphology of *K. gyiorum* on blood agar (**A**) and MacConkey agar (**B**) after 24 h of incubation at 35 °C in 5% CO_2.

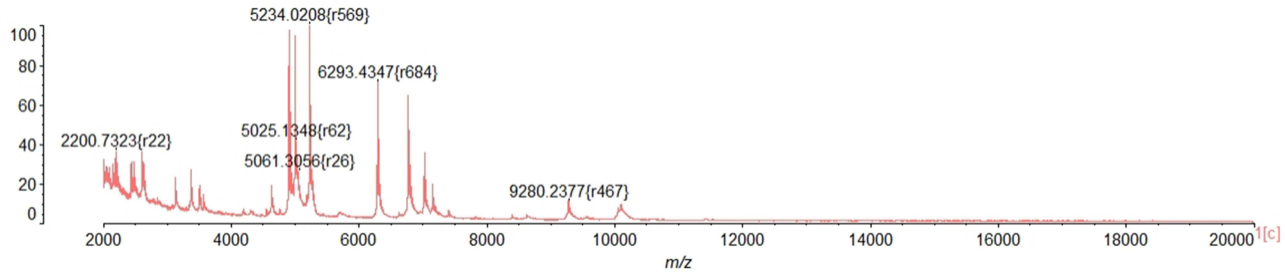

Figure 3 Mass spectrometry image of *K. gyiorum*. r: resolution ratio; red 1[c]: the position of the *K. gyiorum* during VITEK MS in the RUO.

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Table 2 Susceptibility Testing Profile of *K.gyorium* Isolation

Susceptibility Test	MIC Value (ug/mL)	Susceptibility Test	MIC value (ug/mL)
Amikacin	≤8	Cefepime	2
Gentamicin	4	Cefoperazone-Sulbactam	1/8
Tobramycin	≤2	Aztreonam	16
Cefazolin	16	Ampicillin–sulbactam	≤4/2
Cefuroxime	≥16	Piperacillin–tazobactam	≤4/4
Cefoxitin	≤4	Trimethoprim-sulfamethoxazole	≤1/19
Ceftazidime	2	Ciprofloxacin	4
Ceftriaxone	2	Levofloxacin	2

Discussion

Chronic osteomyelitis is a persistent infection of the bone and bone marrow caused by bacteria, mycobacteria, or fungi.[8] It typically follows a relapsing course over months or years, and its management generally requires combination of surgical intervention and prolonged pathogen-directed antibiotic therapy.[8] The patient described in this case had a history of recurrent purulent discharge and fluid leakage from the right lower leg for more than ten years. Although empirical antibiotic regimens administered at other institutions had previously provided symptomatic relief, his condition deteriorated in the two months prior to admission, with increased ulceration and purulent secretion that responded poorly to further antibiotic treatment. Following the identification of *K. gyiorum* was at our hospital, targeted therapy with with Ceftriaxoneled to clinical improvement. Telephone follow-up confirmed that the patient remained in stable condition, underscoring the importance of accurate microbiological diagnosis in the management of chronic osteomyelitis.

We report a case of a cerebral infarction with left limb paralysis, complicated by chronic osteomyelitis due to monomicrobial *K. gyiorum* infection. A PubMed literature review summarized in Table 3 revealed seven articles (10 cases) reporting on suppurative otitis media, four reporting leg infections, two articles (three cases) involving pulmonary infections, and one each documenting urinary tract infection and dermatitis. Among the 19 reported cases, 11 were polymicrobial infections and six were monomicrobial; the present case also represents a monomicrobial infection. Fifteen patients improved following treatment. Literature analysis suggests that risk factors for *K. gyiorum* infection include underlying diseases, prolonged chronic illnesses (some even exceeding ten years), immunocompromised status, repeated hospitalizations, and invasive or traumatic procedures such as surgery or ventilator use.[2–6,9–17] To our knowledge, this is one of the very few reported cases of chronic osteomyelitis caused solely by *K. gyorium* in a patient with post-stroke limb paralysis.

Conventional biochemical identification systems frequently fail to correctly identify *K. gyiorum*. Consistent with reports by Mwalutende et al,[9] Kim et al,[12] and Ogawa et al,[6] the VITEK 2XL GN card (bioMerieux, France) did not yield a definitive identification despite eight positive reactions, likely due to the absence of this species in the system's database. Similarly, Pence et al noted misidentification of *K. gyiorum* by the RapID NF plus assay (Thermo Fisher Scientific,Lenexa, KS) as *Pseudomonas oryzihabitans, Burkholderia cepacia*, or *Acinetobacter spp*. and by the API 20NE system (bioMérieux) as *Alcaligenes faecalis* or *Acinetobacter baumannii/calcoaceticus*.[3] In the present case, VITEK MS in IVD mode also failed to identify the isolate, whereas RUO mode provided correct identification, suggesting limited database coverage in the IVD database. The isolate was successfully identified as *K. gyiorum* by MALDI-TOF MS, consistent with previous reports. These findings highlight the value of advanced technologies such as MALDI-TOF MS, VITEK MS (RUO mode), and 16S rRNA gene sequencingin improving the detection and understanding of rare pathogens like *K. gyiorum*.

The isolate was sensitive to cephalosporins, penicillins, aminoglycosides, and quinolones, in agreement with most published data. However, resistance to ciprofloxacin, gentamicin, and trimethoprim/sulfamethoxazole has been reported in some strains.[2–5,9–11] Although *K. gyiorum* often shows in vitro susceptibility to multiple drug classes, its pathogenicity may be influenced by virulence factors. Li Y et al identified 326 potential virulence factors in strain SWMUKG01, including genes associated with surface polysaccharides, flagella, pili, iron acquisition, secretion systems, two-

Table 3 Clinical Features of Reported Cases of *K. Gyorium*. (Source: PubMed, Medline)

Year	Authors	Age(y)/ Gender	Basic Disease	Diagnosis	Source	Identification Method	Isolated Species	Infection Type	Treatment	Sensitive	Resistant	Outcome
2024[2]	AlSunbul NFl et al	13/Female	Cerebral palsy	Chronic perichondritis of the ear	Ear discharge	MicroScan/VITEK MS	*K. gyorium*	Unknown	CIP/LEV	TZP[a]/IPM[b]/MEM[c]/CAZ[d]	AMK[e]/GEN[f]/TOB[g]/AMP[h]/ AZM[i]/CZO[j]/CTX[k]/FOX[l]/ CIP[m]/SXT[n]	Unknown
2018[9]	Kim JH et al	51/Female	Canal wall-down mastoidectomies	Chronic otitis media	Ear discharge	MALDI-TOF MS/ 16S rRNA/gyrB gene	*K. gyorium*	Single	Cefcapene	AMK/ATM[o]/FEP[p]/CTX/ CAZ/GEN/LEV[q]/TZP/IPM/ MEM/	CIP	Recovered
2018[10]	Özcan N et al	21/Female	Ear discharge and hearing loss	Chronic otitis media	Ear discharge	MALDI-TOF MS	*K. gyorium*	Single	CIP/CRO[r]	AMK/IPM/MEM/TZP	GEN/CIP/CAZ	Recovered
		21/Male	Purulent ear discharge	Chronic otitis media	Ear discharge	MALDI-TOF MS	*K. gyorium*	Single	CIP/CRO	AMK/CRO/CTX/CAZ/CIP	GEN/SXT	Recovered
		27/Male	Hearing loss and ear drainage	Chronic otitis media	Ear discharge	MALDI-TOF MS	*M. morganii and K. gyiorum*	Mixed	CRO/CIP/MFX[s]	AMK/CRO/CTX/CAZ/ MEM	GEN/SXT/CIP	Recovered
2015[11]	Uysal EB et al	25/Male	Otorrhea	Chronic otitis media	Ear discharge	MALDI-TOF MS/ 16S rRNA	*P. aeruginosa and K. gyiorum*	Mixed	IPM	AMK/FEP/CAZ/IPM/MEM/ TZP	CIP/ATM/SXT	Recovered
2014[12]	Mwalutende A et al	53/Male	Ear discharge	Chronic otitis media	Ear discharge	MALDI-TOF MS	*P. mirabilis and K. gyiorum*	Mixed	CIP	CTX/IPM/MEM/CAZ/GEN	CIP(I[w])	Recovered
		33/Male	Ear discharge	Chronic otitis media	Ear discharge	MALDI-TOF MS	S *aureus, E. coli and K. gyiorum*	Mixed	CIP	CTX/IPM/MEM/CAZ/GEN	CIP(I)	Recovered
2012[13]	Almuzara MN et al	16/Male	Otitis media	Chronic otitis media	Purulent discharge	16S rRNA and gyrB genes	*K. gyorium*	Single	CRO/CIP/AMC	Aminoglycosides, CIP, broad-spectrum Cephalosporins	/	Recovered
2013[3]	Pence MA et al	55/Male	Canal wall-down mastoidectomies	Chronic otitis media	Ear discharge	MALDI-TOF MS/ 16S rRNA	*K. gyorium*	Single	SXT	FEP/GEN/MEM/TZP/SXT	CIP	Recovered
		54/Female	Morbidly obesity	Chronic leg wound	Wound secretion	MALDI-TOF MS/ 16S rRNA	*M. morganii and K. gyiorum*	Mixed	CIP	FEP/GEN/MEM/TZP/SXT	CIP	Unknown
2017[14]	Baran I et al	47/Male	Buerger's disease	Chronic foot and ankle infection	Wound secretion	16S rRNA	*P. vulgaris, E. cloacae, M. morganii and K. gyiorum*	Mixed	SAM[u]/CIP	AMK/ATM/CAZ/CIP/GEN/ IPM/MEM/TZP/SXT	Colistin	Recovered
2015[15]	Greninger et al al	57/Male	Diabetes and osteomyelitis	Leg ulcer	Secretion	MALDI-TOF MS/ 16S rRNA	*K. gyorium*	Unknown	Unknown	Unknown	Unknown	Unknown
2015[4]	Bostwick AD et al	69/Female	Schizophrenia	Chronic lower extremity ulcers	Blood	16S rRNA	*K. gyorium*	Single	CIP/CLI[v]	CRO/SXT	CAZ/FEP/TZP/CIP/CTX(I)	Recovered
2014[16]	Deutscher M et al	63/Female	Chronic respiratory failure/end stage renal disease	Tracheobronchitis	Bronchoalveolar Lavage	MALDI-TOF MS/ 16S rRNA	*P. aeruginosa, S. maltophilia and K. gyorium*	Mixed	TZP	AMK/GEN/TOB/FEP/TZP/ SXT	CIP(I)	Death
2023[5]	Sun Y et al	92/Female	Alzheimer's disease	Pulmonary infection	Sputum	MALDI-TOF MS/ 16S rRNA	*K. gyiorum and P. aeruginosa*	Mixed	Traditional chinese and TZP	TZP/ATM/IPM/MEM/TOB / AMK/SXT	CIP/LEV	Recovered
		78/Male	Parkinson's disease	Pulmonary infection	Sputum	MALDI-TOF MS/ 16S rRNA	*K. gyiorum, C. koseri and Morganella*	Mixed	Traditional chinese			Recovered
2016[6]	Ogawa Y et al	82/Male	Bladder cancer	Percutaneous nephrostomy	Urine	MALDI-TOF MS/ 16S rRNA	*P. vulgaris and K. gyiorum*	Mixed	LEV	AMP/AMC[t]/TZP/CTX/ CAZ/FEP/IPM/MEM/LEV/ GEN/AMK	/	Recovered
2021[17]	Kitagawa D et al	48/Male	Swelling of the lower legs	Congestive dermatitis	Granulation tissue	16S rRNA	*B. trematum and K. gyiorum*	Mixed	LEV/MEM	TZP/CAZ/FEP/ATM/IPM/ MEM/AMK/GEN/TOB/SXT	CIP/LEV	Recovered

Notes: [a]:Piperacillin-Tazobactam; [b]:Imipenem; [c]:Meropenem; [d]:Ceftazidime; [e]:Amikacin; [f]:Gentamicin; [g]:Tobramycin; [h]:Ampicillin; [i]:Azithromycin; [j]:Cefazolin; [k]:Cefuroxime; [l]:Cefoxitin; [m]:Ciprofloxacin; [n]:Trimethoprim/Sulfamethoxazole; [o]: Aztreonam; [p]:Cefepime; [q]:Levofloxacin; [r]:Cefotaxime; [s]:Moxifloxacin; [t]:Amoxicillin-Clavulanate; [u]:Ampicillin-Sulbactam; [v]:Clindamycin; [w]:Intermediate.

component systems, and efflux pumps.[18] Nevertheless, the limited number of reported cases makes it difficult to fully assess the clinical significance and pathogenic mechanisms of *K. gyiorum*, warranting further study.

Conclusion

In summary, chronic osteomyelitis is a recalcitrant infection characterized by prolonged duration and a tendency to relapse, often necessitating combined medical and surgical therapy. Optimal antimicrobial selection should be guided by identification and susceptibility testing of the causative pathogen.[8] In this case, we successfully identified *K. gyiorum* using VITEK MS in RUO mode and effectively treated the infection with ceftriaxone in a patient with cerebral infarction and limb paralysis. Technologies such as VITEK MS, MALDI-TOF MS, and 16S rRNA sequencing play crucial roles in recognizing rare pathogens from clinical specimens. Future studies should focus on elucidating the virulence mechanisms and pathogenic potential of this emerging organism.

Ethics Approval and Consent to Participate

Ethical approval was not required for this case report in accordance with the local guidelines. Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Consent for Publication

Written informed consent was obtained from the patient for the publication of this case report.

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Disclosure

The authors report no conflicts of interest in this work.

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