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Case Report

Two cases of chronic suppurative otitis media caused by *Kerstersia gyiorum* in Tanzania: is it an underappreciated pathogen in chronic otitis media?

Alfred Mwalutende [a], Stephen E. Mshana [b], Mariam M. Mirambo [b,*], Martha F. Mushi [b], Philipo L. Chalya [a], Japhet M. Giliyoma [a], Wilhelm Schneiderhan [c], Ortrud Zimmermann [c], Uwe Groß [c]

[a] *Department of Surgery, Weill Bugando School of Medicine, Mwanza, Tanzania*
[b] *Department of Microbiology and Immunology, Weill Bugando School of Medicine, CUHAS-Bugando, PO Box 1464, Mwanza, Tanzania*
[c] *Institute of Medical Microbiology, University Medical Centre, Göttingen, Germany*

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SUMMARY

Two cases of mixed infection involving *Kerstersia gyiorum* causing chronic suppurative otitis media (CSOM) have been reported worldwide. We report, for the first time, two cases of CSOM due to mixed infections involving *K. gyiorum* in adults in Africa. Both isolates were intermediate susceptible to ciprofloxacin based on European Committee on Antimicrobial Susceptibility Testing (EUCAST) breakpoints.

1. Introduction

Chronic suppurative otitis media (CSOM) is defined as ear discharge/otorrhea for more than 6 weeks in the presence of tympanic membrane perforation. The aetiological pattern shows mixed infections of Gram-negative, Gram-positive, aerobe, and anaerobe bacteria. Several studies worldwide have reported that the most common microorganisms isolated are *Pseudomonas spp* and *Staphylococcus aureus*, followed by Gram-negative bacteria such as *Proteus spp*, *Klebsiella spp*, *Escherichia spp*, and *Haemophilus influenzae*.[1] The most frequently isolated anaerobic organisms are *Bacteroides spp* and *Fusobacterium spp*.

Mixed infections have generally been observed in the pathogenesis of CSOM. The role of some bacteria in the aetiology of CSOM has not been well studied.[1] Recently, newly discovered pathogens such as *Kerstersia gyiorum* have been found to be involved.[2,3] The genus *Kerstersia* was first identified in 2003, about 11 years ago, by Coenye et al.[4] The word 'gyiorum', meaning 'from the limbs', was given as a species name since the organism was isolated from wound swabs from limbs. Since then, only two cases of this organism causing CSOM have been reported worldwide. We report, for the first time, two cases of CSOM caused by this organism occurring in one centre in Africa.

2. Case reports

2.1. Case 1

The first patient was a male aged 53 years from rural Mwanza. He reported having a left ear discharge for the past 2.6 years without any other symptoms. The patient had been a smoker and alcoholic for a long time. He was non-diabetic and his HIV status was not known; after counselling, he refused HIV testing.

On examination the patient was found to have conductive hearing loss. The patient reported having used various ear drops, including traditional medicine, without success. He could not remember the types of ear drops used. An ear swab was taken.

On Gram staining, the sample revealed Gram-negative rods and few polymorphonuclear cells. Significant growth of two morphologically different non-lactose fermenter colonies was observed on MacConkey agar. The first were large colourless colonies that were indole-negative, urease-positive, and swarming on blood agar and

* Corresponding author.
E-mail address: mmmirambo@gmail.com (M.M. Mirambo).

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produced hydrogen sulphide; these were identified as *Proteus mirabilis*. The second were medium-sized, light purple in colour, oxidase-negative, indole-negative, catalase-positive, and non-motile. No definitive identification was reached for the second colonies using these in-house biochemical tests. Further testing using a VITEK 2 Gram-negative identification (GNI) card (bioMérieux, Durham, NC, USA) gave a negative result. The colonies were later identified as *K. gyiorum* by matrix-assisted laser desorption/ionization time-of-flight mass spectrometry (MALDI-TOF MS) (Bruker MALDI BioTyper system software version 3.1).

Susceptibility testing was done using VITEK 2 (bioMérieux, France). The *P. mirabilis* was sensitive to ampicillin/sulbactam, piperacillin/tazobactam, cefotaxime, ceftazidime, ciprofloxacin, trimethoprim/sulfamethoxazole, gentamicin, imipenem, and meropenem, while the *K. gyiorum* had intermediate sensitivity to ciprofloxacin (1 μg/ml) and full sensitivity to piperacillin, cefotaxime, ceftazidime, gentamicin, imipenem, meropenem, and moxifloxacin, using the European Committee on Antimicrobial Susceptibility Testing (EUCAST) non-species-related breakpoints. The ciprofloxacin minimum inhibitory concentration (MIC) of 1 μg/ml is considered susceptible using Clinical and Laboratory Standards Institute (CLSI) breakpoints.

The patient was treated with ciprofloxacin ear drops, which were initiated before culture and sensitivity results were obtained. He was scheduled for follow-up 3, 5, and 7 weeks later, but did not return for these scheduled visits. On telephone consultation, he reported doing fine and the drainage had stopped after 10 weeks of treatment. No further complications were noted. After 12 weeks, he reported doing fine without any complications.

2.2. Case 2

The second patient was a male aged 33 years, not related to the patient in case 1 above, who presented with an ear discharge of 1.6 years duration involving the right ear. This patient was seen 3 months after the first case. The patient was a non-smoker and was non-diabetic and HIV-negative. He did not have a history of alcoholism. On examination, conductive hearing loss was found. The patient had used chloramphenicol ear drops several times without success. An ear swab was taken.

On Gram staining, Gram-negative rods mixed with a few Gram-positive cocci were seen. Significant growth of three different colonies was seen on blood agar. Beta-hemolytic golden yellow colonies were identified as *S. aureus* using DNAse and the coagulase test. Significant growth of lactose fermenter and non-lactose fermenter colonies was seen on MacConkey agar. The lactose fermenter colonies were identified as *Escherichia coli* using an in-house biochemical test, while no clear identification was obtained for the non-lactose fermenter colonies. The non-lactose fermenter colonies were oxidase-negative, indole-negative, catalase-positive, and non-motile. As for case 1, further identification using a VITEK 2 GNI card (bioMérieux) gave a negative result. The colonies were later identified as *K. gyiorum* using MALDI-TOF MS (Bruker MALDI BioTyper system software version 3.1).

On VITEK 2 susceptibility testing, the *S. aureus* was resistant to ampicillin, trimethoprim/sulfamethoxazole, rifampin, and tetracycline and was methicillin-sensitive. The *E. coli* had an extended-spectrum beta-lactamase (ESBL) phenotype and was resistant to ampicillin, sulbactam, piperacillin/tazobactam, cefotaxime, ceftazidime, ciprofloxacin, and trimethoprim/sulfamethoxazole, and was intermediate resistant to gentamicin and sensitive to imipenem and meropenem. The sensitivity test results for this isolate of *K. gyiorum* were identical to those of case 1.

Ciprofloxacin ear drops were started before culture results were obtained and the patient was scheduled for follow-up 3, 5, and 7 weeks later. He was doing fine at the subsequent visits and the drainage had stopped.

3. Discussion

Information on the aetiology of CSOM is limited in Africa; this may be attributed to poor diagnostic facilities. There is the possibility that various pathogens are involved in the pathogenesis of CSOM in Africa. In Africa, including Tanzania, most clinical microbiology laboratories have limited identification capacity. This might result in misidentification or failure to identify common isolates causing certain pathologies. The description of these two cases in Tanzania with microbial identification confirmed using MALDI-TOF MS shows the importance of advanced technologies in the identification of novel and rare pathogens from clinical specimens. In developing countries where few biochemical tests are used for identification, this organism might have been misidentified. *K. gyiorum* strains exhibit some features of *Acinetobacter spp*, which are also non-fermentative Gram-negative rods and oxidase-negative. The organism is also closely related to other members of the genera *Alcaligenes*[5] and *Achromobacter*.

K. gyiorum may represent an underappreciated cause of CSOM worldwide as evidenced by the fact that, in the past 2 years, two cases of CSOM caused by *K. gyiorum* have been reported, one in the USA and one in South America.[2,3] As previously documented,[2,3] both patients had CSOM involving *K. gyiorum* mixed with other organisms. It is likely that *K. gyiorum* has an affinity towards causing infections in patients with chronic mixed infections. However, little is known regarding how the organism contributes to the chronicity of the infected sites, and its virulence factors are yet to be identified.

One of the cases presented here was a chronic smoker and an alcoholic; these factors were reported in one of the previous cases.[3] More studies are needed to establish the role of these factors in the pathogenesis of this particular infection. These two isolates had intermediate resistance to ciprofloxacin based on EUCAST breakpoints; one of the two previously reported isolates from CSOM was resistant to ciprofloxacin based on CLSI breakpoints[3] and the other was fully susceptible to ciprofloxacin based on CLSI breakpoints.[2]

We have reported these rare cases to alert clinicians to the fact that *K. gyiorum* could contribute significantly as a possible aetiology of CSOM in adults. The identification of this pathogen requires advanced techniques, which are limited in developing countries. There is a need to establish simple molecular techniques for the identification of bacteria in developing countries. This will reduce misidentification of common pathogens involved in various pathologies in resource-limited countries.

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