ISSN 2607-9917 www.bdd.rdplf.org Volume 6, n° 3, november 2023 https://doi.org/10.25796/bdd.v6i3.80703

Bulletin de la Dialyse à Domicile
Home Dialysis Bulletin (BDD)

International bilingual journal for the exchange of knowledge and experience in home dialysis.

(English edition)

Sphingobium yanoikuyae peritonitis in peritoneal dialysis: a case report

(Péritonite à *Sphingobium yanoikuyae* en dialyse péritonéale : à propos d'un cas)

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To cite: Mouradi S, Motte G, Torner S, Lebugle P, Petitboulanger N, Bemmerzouk A, Charles P-Y. Sphingobium yanoikuyae peritonitis in peritoneal dialysis: a case report. Bull Dial Domic [Internet]. 6(3):123-7. Available from: https://doi.org/10.25796/bdd.v6i3.80703

Note : ce texte est disponible en Français à la même adresse url : https://doi.org/10.25796/bdd.v6i3.80703

Summary

We report a rare clinical case of infection with the bacterium *Sphingobium yanoikuyae* in an 87-year-old patient undergoing peritoneal dialysis for end-stage renal failure. *Sphingobium yanoikuyae* is an aerobic, gram-negative bacterium known for its ability to degrade polycyclic aromatic hydrocarbons and its bioremediation potential. As a member of the *Sphingomonadaceae* family, it has been identified in a variety of environments, including dialysis equipment.

After starting peritoneal dialysis, the patient developed an infectious syndrome. Bacteriological analysis of the peritoneal effluent revealed the presence of *Sphingobium yanoikuyae* in the dialysate. Appropriate antibiotic therapy with meropenem was instituted as soon as the Sphingobium antibiogram was obtained. A second bacterium, *Sphingomonas sp*., was also subsequently identified (resistant to meropenem). Given the favorable clinicobiological course, only *Sphingobium* was considered to be responsible for the disease.

This is the first known case of human infection with *Sphingobium yanoikuyae*, and the third case of infection with a *Sphingobium* species in a peritoneal dialysis setting. The low prevalence of this germ in human infections suggests a low virulence of this bacterium. This nevertheless highlights the potential risk of nosocomial infections linked to this family of bacteria. This germ has shown antibiotic resistance, raising concerns about anti-infective resistance in opportunistic bacteria such as *Sphingomonadaceae*.

This case adds to our knowledge of rare, antibiotic-resistant infections in the hospital setting, particularly in vulnerable patients treated with peritoneal dialysis.

Keywords: peritoneal dialysis, peritonitis, Sphingomonadaceae, Sphingobium yanoikuyae, meropenem.

Résumé

Nous rapportons un cas clinique rare d'infection par la bactérie *Sphingobium yanoikuyae* chez un patient de 87 ans traité par dialyse péritonéale pour insuffisance rénale terminale. *Sphingobium yanoikuyae* est une bactérie aérobie, gram-négative, connue pour sa capacité à dégrader les hydrocarbures aromatiques polycycliques et son potentiel en bioremédiation. Elle fait partie de la famille des *Sphingomonadaceae*, identifiée dans divers environnements, y compris les équipements de dialyse.

Après avoir commencé la dialyse péritonéale, le patient a développé un syndrome infectieux. L'analyse bactériologique de l'effluent péritonéal a mis en évidence la présence de *Sphingobium yanoikuyae* dans le dialysat. Une antibiothérapie adaptée par meropenem a été institué dès obtention de l'antibiogramme du Sphyngobium. Une seconde bactérie, Shingomonas sp était également identifiée dans les suites (résistant au meropenem). Du fait d'une évolution clinicobiologique favorable, seul le *sphingobium* a été retenu responsable de l'atteinte.

Ce cas est le premier connu d'infection humaine à *Sphingobium yanoikuyae* et le troisième cas d'infection par une espèce de *Sphingobium* en contexte de dialyse péritonéale. La faible prévalence de ce germe dans les infections humaines suggère une faible virulence de cette bactérie. Cela met néanmoins en évidence le risque potentiel des infections nosocomiales liées à cette famille de bactéries. Ce germe a montré une résistance aux antibiotiques, ce qui soulève des préoccupations sur la résistance aux anti-infectieux chez les bactéries opportunistes comme les *Sphingomonadaceae*.

Ce cas ajoute aux connaissances sur les infections rares et résistantes aux antibiotiques en milieu hospitalier, en particulier chez les patients vulnérables traités par dialyse péritonéale.

Mots clés : péritonite, dialyse péritonéale, Sphingomonadaceae, Sphingobium yanoikuyae, meropenem.

journal officiel du Registre de Dialyse Péritonéale de Langue Française **RDPLF** www.rdplf.org

www.bdd.rdplf.org Volume 6, n° 3, november 2023
https://doi.org/10.25796/bdd.v6i3.80703
ISSN 2607-9917

Introduction

Sphingobium yanoikuyae is a short, rod-shaped, strictly aerobic, gram-negative, non-motile, non-spore-forming bacterium of the *Sphingomonadaceae* family. It is a hydrophilic bacterium capable of forming biofilms.
Bacteria of the *Sphingomonadaceae* family have been described in a variety of terrestrial and aquatic environments, including dialysis and water supply equipment (1). *Sphingobium yanoikuyae* is implicated in the degradation of polycyclic aromatic hydrocarbons. This could be exploited in the development of bioremediation technologies (2).

Peritonitis in peritoneal dialysis is associated with significant morbidity and mortality, and in 4% of cases it can lead to death, damage to the peritoneal membrane, and withdrawal of the dialysis catheter, with temporary or permanent recourse to hemodialysis.

Evolution of the taxonomy of the genera Sphingomonas and Sphingobium

Also belonging to the *Sphingomonadaceae* family, the Sphingomonas genus was described in 1990 by Yabuuchi et al. (3), identifying *Sphingomonas paucimobilis* as the type specie, and new species in the genus including *Sphingomonas yanoikuyae*.

The first description of the *Sphingobium* genus was made by Takeuchi et al. in 2001 (4), distinguishing it from *Sphingomonas* after phylogenetic analyses of 16S rRNA gene sequences. *Sphingobium yanoikuyae* is the type species of the genus *Sphingobium* (5).

Sphingomonas yanoikuyae is the basionym of Sphingobium yanoikuyae, although nomenclature defines them as two distinct species.

While a dozen human Sphingomonas infections of different types have been described (6,7), only two cases of human infections caused by *Sphingobium* species have been reported. These latter cases involved infection of peritoneal dialysis fluid:
- *Sphingobium* olei peritonitis (8)
- *Sphingobium* lactosutens peritonitis (9)

Clinical observation

We describe here an 87-year-old patient followed in nephrology for end-stage renal failure of diabetic and vascular origin. He has been treated with peritoneal dialysis since the end of May 2023, after catheter placement in mid-April. The patient was trained and then educated in continuous ambulatory peritoneal dialysis (CAPD). After this education phase, the patient was able to carry out his care independently at home. The peritoneal dialysis protocol included two isotonic bags during the day and one Icodextrin bag at night, six days a week.

A few days after initiation of peritoneal dialysis, the patient presented a deterioration in general condition, accompanied by diabetic imbalance, warranting hospitalization due to the intensity of the anorexia.
Clinical examination was unremarkable except for constipation for three days.

journal officiel du Registre de Dialyse Péritonéale de Langue Française RDPLF www.rdplf.org

www.bdd.rdplf.org Volume 6, n° 3, november 2023
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Initial laboratory workup revealed an infectious syndrome, with C-reactive protein at 223 mg/l, hyperleukocytosis at 10.9 G/L, and slightly increased procalcitonin at 0.67 ng/ml.

On 06/06/2023, analysis of the dialysate showed a cloudy appearance, with a negative blood agar culture on the first bag. Nevertheless, cellular analysis of this first dialysate sample showed an increase in leukocytes to 750/mm3, including 472 neutrophils per mm3, with numerous mesothelial cells and macrophages.
While analysis of the second bag showed no significant increase in leukocytes (48/mm3 including 13% neutrophils, 43% lymphocytes, 25% histiocytic cells), a culture on blood agar identified in 36 hours:

- Numerous *Sphingobium yanoikuyae*:
    - Sensitive: meropenem, ceftazidime + avibactam
    - Intermediate: ceftazidime, ciprofloxacin
    - Resistant: ceftriaxone, amoxicillin
- Fairly numerous *Sphingomonas sp*.:
    - Sensitive: amoxicillin and ciprofloxacin
    - Resistant: meropenem, ceftazidime, ceftriaxone, ceftazidime + avibactam

These two bacteria are closely related in genus but remain distinct.

On both bags, neither direct examination nor blood cultures on dialysate taken simultaneously identified any germs.

The patient was initially treated with vancomycin 1 gram and ceftriaxone 2 grams IV probabilistic therapy. As soon as the *Sphingobium antibiogram* was obtained, the patient was switched to meropenem 1 gram/d IV, then intraperitoneally with 6-hour stasis for a total of three weeks. A second bacterium, *Sphingomonas s*p. (resistant to MEROPENEM), was also identified later. Given the favorable clinicobiological course, only *Sphingobium* was considered to be responsible for the disease.

During hospitalization, the patient developed a peripheral a frigore facial paralysis, with sudden deafness, which did not regress. No etiology was identified for these symptoms. There was possible neurotoxicity of meropenem (10,11) but no plasma meropenem assays were performed to support this hypothesis. Nevertheless, this prescription was in line with antibiotic therapy recommendations for dialysis patients.

Bacteriological analysis of the dialysate came back sterile after 48 hours of treatment, with an improvement in clinical condition and a favorable evolution of biological inflammatory markers.

Conclusion

This clinical case reports the first case of human infection with *Sphingobium yanoikuyae*, and it is the third case of *Sphingobium* infection, all in the context of peritoneal dialysis fluid infection.

This picture of pauci-symptomatic peritonitis, which progressed favorably under antibiotic therapy targeting *Sphingobium yanoikuyae* and not covering the susceptibility of *Sphingomonas sp*., is in favor of low virulence in our patient.

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www.bdd.rdplf.org Volume 6, n° 3, november 2023
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However, although rare, descriptions in the literature of infections with *Sphingomonas* species suggest an increase in the incidence of nosocomial infections with the *Sphingomonadaceae* family, whose resistance profile is still poorly studied (nevertheless, an intrinsic resistance to colistin and an extended resistance to beta-lactams have been described). Their omnipresence in the environment and their adaptability constitute a natural reservoir of antibiotic resistance, which is potentially dangerous for humans due to their opportunistic nature (12).

Ethical considerations

The patient has given his consent for the publication of this clinical case.

Role des auteurs

Sara Mouradi: patient management, article writing
Gérard Motte: proofreading the article
Stéphane Torner: proofreading the article
Pierre Lebugle: patient management
Nelly Petitboulanger: bacteriological analysis of samples
Aziz Bemmerzouk: final proofreading of the article
Pierre-Yves Charles: bibliography, article writing, proofreading, final corrections

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Received 2023-07-19, accepted after peer reviewing 2023-09-25, published 2023-11-23

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