A case of *Sphingomonas paucimobilis* causing peritoneal dialysis-associated peritonitis and review of the literature

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**Abstract**

**Background:** Peritoneal dialysis (PD)-associated peritonitis caused by *Sphingomonas paucimobilis* (*S. paucimobilis*) is very rare, and most of the characteristics of such cases are still unknown.

**Case presentation:** An 80-year-old Japanese woman on PD was diagnosed with PD-associated peritonitis and received ceftazidime and cefazolin. The number of cells in the peritoneal dialysate decreased quickly. However, because *S. paucimobilis* was detected, the antibiotic was changed to meropenem according to the susceptibility test results. She was treated with meropenem for two weeks and discharged. After 21 days, she was hospitalized for relapsing peritonitis. *S. paucimobilis* was detected again, and improvement after the administration of meropenem was poor, eventually resulting in catheter removal.

**Conclusions:** *S. paucimobilis* may be resistant to empirical antibiotics; furthermore, catheter removal may still be required, even with sensitive-antibiotic treatment.

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*S. paucimobilis* is a non-fermentative Gram-negative bacillus that is widely distributed in nature and is also present in the hospital environment [1, 2]. *S. paucimobilis* rarely infects humans, but when it does, it is suspected to cause meningitis, urinary tract infection, endophthalmitis, splenic abscess, arthritis, osteomyelitis, empyema, pneumonia, and catheter-related infection [2–4]. It has been reported that contaminated solutions, such as distilled water, haemodialysis fluid and sterile drug solutions, cause bacteremia and sepsis [2]. The pathogenicity of *S. paucimobilis* is considered to be low because the prognosis is generally good despite inappropriate treatment [3]. *S. paucimobilis* infection is most likely to occur in patients complicated with underlying diseases, such as malignant carcinoma, immunodeficiency, or diabetes [3, 4]. The incidence of peritoneal dialysis (PD)-associated peritonitis has decreased due to technological progress [5], but it remains an important complication and a major cause of PD withdrawal [6]. In this report, we present the case of a woman with PD-associated peritonitis caused by *S. paucimobilis* who had to undergo catheter removal and PD withdrawal. Given that studies on *S. paucimobilis*-related peritonitis are rarely reported, this is considered a valuable case.

**Case presentation**

An 80-year-old Japanese woman developed end-stage renal disease due to nephrosclerosis 2 years previously and started maintenance haemodialysis. Six months later, PD, instead of haemodialysis, was initiated due to remarkably low left heart function. She underwent three-cuff Swan neck catheter implantation. She used an automated connecting device with ultraviolet light undergoing PD. She visited our hospital after experiencing discomfort;
she was admitted because of cloudy dialysate and mild abdominal pain starting on the previous day. The dialysis effluent white cell count was 4875/μL, with a predominance of neutrophils (86%), and she was diagnosed with PD-associated peritonitis. On admission, her body temperature was 36.4 °C, blood pressure was 133/71 mmHg, heart rate was 70/min, height was 155 cm, and weight was 49.9 kg. Only mild abdominal tenderness was noted without muscular defence or rebound tenderness. Diarrhoea was not present. No apparent signs of infection at the exit or in the tunnel were observed. Laboratory findings on admission are summarized in Table 1. Her haemoglobin was 12.2 g/dL, white blood cell (WBC) count was 5600/μL, C-reactive protein was 3.20 mg/dL (normal < 0.3 mg/dL), and albumin was 2.6 g/dL. Blood culture was negative. When 1 g of ceftazidime and 1 g of cefazolin were administered intraperitoneally daily, the dialysis effluent white cell count decreased rapidly. On the fourth day, Gram-negative rods were detected in the peritoneal fluid culture, so only ceftazidime was administered. The causative organism was subsequently identified as *S. paucimobilis*. Given that this organism was resistant to ceftazidime and sensitive to meropenem (Table 2), antibiotic treatment was changed to 0.5 g of intravenous meropenem per day on the seventh day. This dosage was administered for 2 weeks. After the antibiotic was changed to meropenem, the dialysis effluent white cell count was continuously greater than 100/μL, but the neutrophil count in the dialysis effluent was decreased. The neutrophil percentage was 3% on the 18th day. Therefore, we considered meropenem to be effective. The patient was discharged on the 21st day (Fig. 1).

However, three weeks later, the dialysate became cloudy again. The dialysis effluent white cell count was 2604/μL, with a predominance of neutrophils (97%). She was admitted a second time for relapsing peritonitis. On the second admission, her body temperature was 36.7 °C, blood pressure was 129/73 mmHg, and heart rate was 65/min. As before, mild tenderness was noted throughout the abdomen, without muscle defence or rebound tenderness. No clear signs of infection at the exit tunnel were observed. The haemoglobin level was 10.4 g/dL, WBC count was 5800/μL, C-reactive protein was 2.27 mg/dL, and albumin was 2.5 g/dL (Table 1). From the first day of the second hospitalization, 0.5 g of intravenous meropenem was administered daily. *S. paucimobilis* was detected again in the peritoneal fluid culture during the second admission, and sensitivity testing indicated that it was sensitive to meropenem (Table 2). On the sixth day of the second hospitalization, the dialysis effluent white cell count was 1376/μL. Due to poor improvement, 15 mg of tobramycin was additionally administered intraperitoneally daily. On the ninth day of the second hospitalization, the dialysis effluent white cell count decreased to 337/μL, but we hypothesized that peritonitis was not controlled, because *S. paucimobilis* was detected again in the peritoneal fluid culture on the 6th day. Furthermore, we were worried that the patient was exhausted. On the next day, the PD catheter was removed (Fig. 1). The culture results of the internal cuff, middle cuff, external cuff, catheter between the internal cuff and middle cuff, catheter between the middle cuff and external cuff, and catheter tip were

| Table 1 Laboratory data |
|-------------------------|
|                         | First admission | Second admission |
| WBC                     | 5600           | 5800           |
| RBC                     | 389 x 10⁴      | 347 x 10⁴      |
| Hb                      | 12.2           | 10.4           |
| Ht                      | 35.1           | 30.2           |
| MCV                     | 90.2           | 87.0           |
| Plt                     | 14.9 x 10⁴     | 18.8 x 10⁴     |
| Cr                      | 6.88           | 6.16           |
| BUN                     | 56.6           | 49.2           |
| BMG                     | 26.6           | 18.8           |
| TP                      | 6.0            | 5.7            |
| Alb                     | 2.6            | 2.5            |
| Na                      | 138            | 137            |
| K                       | 3.5            | 3.2            |
| Cl                      | 101            | 99             |
| Ca                      | 8.4            | 8.0            |
| P                       | 4.6            | 3.8            |
| Glu                     | 131            | 105            |
| ALT                     | 9              | 10             |
| AST                     | 7              | 6              |
| LDH                     | 205            | 236            |
| ALP                     | 384            | 265            |
| γGTP                    | 19             | 19             |
| T.Bil                   | 0.4            | 0.6            |
| BNP                     | NA             | 333.6          |
| CRP                     | 3.2            | 2.27           |
| Blood culture           | –              | NA             |
| Peritoneal WBC          | 4875           | 2604           |
| Neu                     | 86             | 97             |
| Lym                     | 6              | 2              |
| Mon                     | 8              | 0              |
| Eos                     | 0              | 1              |

WBC, WHITE BLOOD cell count; RBC, red blood cell count; Hb, haemoglobin; Ht, haematocrit; MCV, mean corpuscular volume; Plt, platelet count; Cr, creatinine; BUN, blood urea nitrogen; BMG, β₂-microglobulin; TP, total protein; Alb, albumin; Glu, glucose; ALT, alanine aminotransferase; AST, aspartate aminotransferase; LDH, lactate dehydrogenase; ALP, alkaline phosphatase; γGTP, γ-glutamyl transpeptidase; T.Bil, total bilirubin; BNP, brain natriuretic peptide; CRP, C-reactive protein; Neu, neutrophil; Lym, lymphocyte; Mon, monocyte; Eos, eosinophil
negative. She transitioned to haemodialysis, continued to receive meropenem for 15 days after surgery and was discharged.

**Discussion and conclusions**

*S. paucimobilis* is an unusual pathogen for PD-associated peritonitis. We report a case of peritonitis due to *S. paucimobilis* that required catheter removal.

Lin et al. reported 42 cases of *S. paucimobilis* bacteraemia [3]. In that study, primary *S. paucimobilis* bacteraemia was found in 35.7% of patients. Catheter-related bloodstream infections were identified in 33.3% of patients, skin and soft tissue infections were identified in 9.5% of patients, pneumonia was identified in 9.5% of patients, urinary tract infections were identified in 4.8% of patients, biliary tract infections were identified in 4.8% of patients, and meningitis was identified in 2.4% of patients. Although three patients experienced septic shock, all 42 patients survived the *S. paucimobilis* bacteraemia episodes. The authors concluded that *S. paucimobilis* exhibited low clinical virulence. *S. paucimobilis* infections other than PD-related peritonitis may not be difficult to treat.

Fourteen cases of PD-associated peritonitis due to *S. paucimobilis* have been reported. The clinical characteristics of this case and the cases reported thus far are summarized in Table 3 [7–19]. The ages of patients ranged from 3.5 to 80 years, and there was no difference in the male-to-female ratio at 7:8. The reports included 2 diabetic patients, 10 nondiabetic patients, and 3 patients with unknown status, and no particular diabetic complications were noted. There were two cases in which the

| Culture of peritoneal dialysate | First admission | Second admission |
|---------------------------------|----------------|-----------------|
| S. paucimobilis                 | Susceptible (≤8) | Susceptible (≤8) |
| Piperacillin                    | Susceptible (≤8) | Susceptible (≤8) |
| Ceftazidime                     | Resistant (>16)  | Resistant (>16)  |
| Cefepime                        | Intermediate (16) | Intermediate (16) |
| Imipenem/cilastatin             | Susceptible (≤1) | Susceptible (≤1) |
| Meropenem                       | Susceptible (≤1) | Susceptible (≤1) |
| Aztreonam                       | Resistant (>16)  | Resistant (>16)  |
| Tazobactam/piperacillin         | Susceptible (≤8) | Susceptible (≤8) |
| Gentamicin                      | Susceptible (≤2) | Susceptible (≤2) |
| Tobramycin                      | Susceptible (≤2) | Susceptible (≤2) |
| Amikacin                        | Susceptible (≤8) | Susceptible (≤8) |
| Minocycline                     | Susceptible (≤2) | Susceptible (≤2) |
| Levofloxacin                    | Resistant (>2)   | Resistant (>2)   |
| Ciprofloxacin                   | Resistant (>2)   | Resistant (>2)   |
| Sulfamethoxazole—trimethoprim   | Susceptible (≤2) | Susceptible (≤2) |

*Table 2* Culture of peritoneal dialysate, susceptibility (minimum inhibitory concentration, μg/mL)

**Fig. 1** Clinical course of the present case
| Case | Year | Age | Gender | DM | Symptoms (first visit) | Susceptible antibiotics | Treatment | Clinical course | Outcome | References |
|------|------|-----|--------|----|------------------------|-------------------------|-----------|----------------|---------|------------|
| 1    | 1984 | 74  | Female | No  | Abdominal pain         | ABPC CBPC CBPC          | ST IP (14 days) | Rapidly improved | Cured   | [7]        |
|      |      |     |        |     | Vomiting               | GM TOB EM TOB ST CP     |           |                 |         |            |
|      |      |     |        |     | Cloudy dialysate       |                         |           |                 |         |            |
| 2    | 1984 | 33  | Female | No  | Abdominal pain         | ABPC CBPC CBPC          | 1. CEZ + TOB IP | 1 Week after treatment | Catheter removed | [7]        |
|      |      |     |        |     | Vomiting               | GM TOB EM TOB ST CP     | 2. ABPC IP    | No3, relapsed    |         |            |
|      |      |     |        |     | Cloudy dialysate       |                         | 3. AMPC orally (5 days) |                 |         |            |
|      |      |     |        |     |                        |                         | 4. After catheter removal, TOB IV |                 |         |            |
| 3    | 1985 | 61  | Male   | No  | Cloudy dialysate       | CXM CAZ Ticarcillin AMK CP | VCM IP + GM IP (duration NR) | NR | Cured | [8]        |
| 4    | 1985 | 50  | Male   | NR  | Cloudy dialysate       | NR                      | 1. CET IP (4 days) | 3 Weeks after treatment | Cured   | [9]        |
|      |      |     |        |     |                        |                         | 2. CET IP (5 days) | No1, first relapsed, after 1 week of treatment no. 3, second relapsed |         |            |
|      |      |     |        |     |                        |                         | 3. CEX orally (14 days) |                 |         |            |
|      |      |     |        |     |                        |                         | 4. TOB IP (14 days) |                 |         |            |
| 5    | 1987 | 65  | Male   | No  | NR                     | MZ CTX                  | 1. VCM (10 days) + TOB (12 days) + ABPC (3 days) | NR | Catheter removed | [10]    |
|      |      |     |        |     |                        |                         | 2. MZ (13 days) + CX (13 days) |                 |         |            |
|      |      |     |        |     |                        |                         | 3. CP (13 days) |                 |         |            |
| 6    | 1988 | 38  | Female | No  | Abdominal discomfort   | CET TOB                | CET IP + TOB IP (duration NR) | 4 days after improvement, relapsed | Catheter removed | [11]    |
|      |      |     |        |     | Nausea                 |                         |                         |                 |         |            |
| 7    | 1990 | 64  | Female | No  | Cloudy dialysate       | Aminoglycosides        | 1. CPFX orally (5 days) | Rapidly improved | Cured | [12]      |
|      |      |     |        |     |                        | ST                     | 2. NTL IP (duration NR) |                 |         |            |
| 8    | 2007 | 51  | Male   | Yes | Abdominal pain         | CAZ CTX CFPM IPM       | 1. CEZ + AMK (14 days) | 12 days after improvement of the first peritonitis due to C. indologenes with treatment of No. 1, developed | Catheter removed | [13]    |
|      |      |     |        |     | Fever                  | SBT/CPZ TAZ/PIPC AMK CPFX | 2. CEZ + CAZ (4 days) |                 |         |            |
|      |      |     |        |     | Cloudy dialysate       |                         |                         |                 |         |            |
| 9    | 2008 | 50  | Male   | NR  | Abdominal pain         | ABPC PIPC IPM SBT/ABPC SBT/CPZ TAZ/PIPC GM LFX ST | 1. VCM IP, single dose | Continued growth of S. paucimobilis despite dialysate without WBCs | Catheter removed | [14]    |
|      |      |     |        |     | Cloudy dialysate       |                         | 2. IPM IV + GM IP (18 days) |                 |         |            |
|      |      |     |        |     |                        |                         | 3. After catheter removal, IPM IV (7 days) |                 |         |            |
| 10   | 2011 | 3.5 | Male   | No  | Abdominal pain         | MEPM AMK TC PL         | 1. AMK IP (4 days) | Rapidly improved | Cured | [15]      |
|      |      |     |        |     | Fever                  |                         | 2. MEPM IV |                 |         |            |
| 11   | 2013 | 63  | Male   | Yes | Abdominal pain         | CAZ CTX IPM MEPM GM MINO CPFX | 1. CEZ + CAZ IP (14 days) | The next day after treatment of No1, relapsed Resistant to CAZ | Catheter removed | [16]    |
|      |      |     |        |     | Cloudy dialysate       |                         | 2. IPM IP |                 |         |            |
| 12   | 2015 | 50  | Female | NR  | Abdominal pain         | CFP MEPM AMK CAM CPFX | 1. VCM IP + CPFX IV (1 day) | Improvement after treatment of No 3 | Cured | [17]      |
|      |      |     |        |     | Vomiting               |                         | 2. TOB IP + CPFX IV (3 days) |                 |         |            |
|      |      |     |        |     | Fever                  |                         | 3. TOB IP + MEPM IV (21 days) |                 |         |            |
### Table 3 (continued)

| Case | Year | Age | Gender | DM | Symptoms (first visit) | Susceptible antibiotics | Treatment | Clinical course | Outcome | References |
|------|------|-----|--------|----|-------------------------|-------------------------|-----------|------------------|---------|------------|
| 13   | 2016 | 35  | Female | No | Abdominal pain          | CTRX CFPM IPM           | 1. VCM IP + CAZ IP (3 days)  
2. CPFX orally + CTRX IP (21 days)  
3. After catheter removal, CPFX orally + CTRX IP (14 days) | 3 days after treatment of No.2, relapsed | Catheter removed | [18]       |
|      |      |     |        |    | Cloudy dialysate        | CPFX LVFX               | 1. VCM IP + CAZ IP (3 days)  
2. CPFX orally + CTRX IP (21 days)  
3. After catheter removal, CPFX orally + CTRX IP (14 days) | 3 days after treatment of No.2, relapsed | Catheter removed | [18]       |
| 14   | 2018 | 63  | Female | No | Abdominal pain          | CAZ AMK GM CPFX         | 1. CAZ IP + VCM IP (3 days)  
2. CAZ IP + AMK IP (21 days) | Rapidly improved  
Cured | [19]       |
|      |      |     |        |    | Vomiting                 |                         |           |                  |         |            |
|      |      |     |        |    | Fever                    |                         |           |                  |         |            |
|      |      |     |        |    | Cloudy dialysate        | Table 2                | 1. CAZ IP (7 days) + CEZ IP (4 days)  
2. MEPM IV (14 days)  
3. MEPM IV (21 days) TOB IP (2 days) | 3 weeks after treatment No.2, relapsed | Catheter removed |           |

DM, diabetes mellitus; ABPC, ampicillin; AMK, amikacin; AMPC, amoxicillin; CAM, clarithromycin; CAZ, ceftazidime; CBPC, carbenicillin; CFPM, cefepime; CPFX, ciprofloxacin; CXM, cefuroxime; CP, chloramphenicol; CEZ, cepalexin; CET, cefalotin; CTRX, ceftriaxone; CTX, cefotaxime; CX, cefoxitin; EM, erythromycin; GM, gentamicin; IPM, imipenem; LVFX, levofloxacin, MEPM, meropenem; MINO, minocycline; MZ, mezlocillin; NTL, netilmicin; PL, polymyxin B; SBT/ABPC, sulbactam/ampicillin; SBT/CPZ, sulbactam/cefoperazone; ST, sulfamethoxazole–trimethoprim; TAZ/PIPC, tazobactam/piperacillin; TC, tetracycline; TOM, tobramycin; VCM, vancomycin; IP, intraperitoneally; IV, intravenously; NR, not reported
and 13.6% were resistant to amikacin [21]. In the pre-
bacteria in reported cases were resistant to cefotaxime,
[15, 20]. However, Bayram et al. reported that 20.0% of
for the treatment of infections caused by this organism
cephalosporin have been suggested as suitable antibiotics
alone and an aminoglycoside plus a third-generation
lead to a refractory infection. Imipenem or meropenem
the appropriate treatment can be delayed, which may
lead to a refractory infection. Imipenem or meropenem
alone and an aminoglycoside plus a third-generation
cephalosporin have been suggested as suitable antibiotics
for the treatment of infections caused by this organism
[15, 20]. However, Bayram et al. reported that 20.0% of
cell count decreased rapidly after ceftazidime and sensitiv-
S. paucimobilis
is extremely rare, but it
described the cases reported thus far. PD-associated
infections due to S. paucimobilis
was sensitive to cefazidime in a few cases. Regarding empirical treatment of
PD-associated peritonitis, selection of third-generation
cephalosporins or aminoglycosides is recommended for
Gram-negative bacteria [6]. However, it should be noted
S. paucimobilis may be resistant to these antibiotics.
In the case reported by Lee et al., the bacterium was ini-
tially sensitive to ceftazidime, and peritonitis improved,
but when it recurred, the bacterium showed resistance to
to ceftazidime, resulting in catheter removal [16]. Therefore,
antibiotic resistance was observed during monotherapy
with ceftazidime. Even if S. paucimobilis is sensitive to
third-generation cephalosporins, the addition of an ami-
glycoside to a third-generation cephalosporin may be
beneficial. The organism responsible for infection of the
patient in the present case was resistant to ceftazidime
sensitive to meropenem. Antibiotic resistance during
monotherapy with meropenem was not noted. However,
because this bacterium was detected during meropenem
administration, the combined use of another antibiotic
(e.g. tobramycin) should have been considered when relapse
occurred.

In this review, 5 of 8 patients with catheter removal,
including the present patient, relapsed after improve-
ment and required catheter removal. Biofilm formation
is one of the causes of relapse. Although the culture
results of the areas between the cuffs and catheter tip
were negative, we suggest that it is possible that a bio-
film was formed on the PD catheter in this patient. Nod-
aira et al. reported that all catheters removed because
of PD-associated peritonitis showed biofilms on elec-
tron microscopy (EM) scanning; however, patients with
catheters removed for other reasons, such as gastroin-
testinal neoplasm or perforation, did not demonstrate
biofilms [22]. EM scanning might have detected a bio-
film in the present case. The administration of merope-
nem was delayed for this patient. Generally, a delay in
starting initial antimicrobial therapy can allow patho-
gens to proliferate, rendering patients less responsive to
treatment. Moreover, biofilms that form due to delayed
treatment initiation might increase the risk of catheter
removal by contributing to the re-development of peri-
onititis after initiation of treatment [23]. If S. paucimo-
bilis is detected and initiation of appropriate treatment
is delayed, careful observation is important even after
treatment with effective antibiotics because of the risk of
relapse.

In this case, the patient was treated with meropenem
for only 2 weeks during the first admission because the
dialysis effluent white cell count decreased rapidly after
to ceftazidime and ceftazidime were administered, and cefta-
zidime was administered for one week. The Interna-
tional Society for Peritoneal Dialysis Guidelines (ISPD
GL) recommend treating Gram-negative bacilli peri-
onititis with effective antibiotics for three weeks [6].
Recent studies have reported success in patients who
were treated with two antibiotics for three weeks [17,
19]. More than half of the previous reports described
failure to eradicate this bacterium, suggesting that
two effective antibiotics are needed for three weeks
(Table 3).

In summary, we treated a patient with PD-associated
peritonitis due to S. paucimobilis
is extremely rare, but it
is important because catheter removal is often required.

Abbreviations
PD: Peritoneal dialysis; S. paucimobilis: Sphingomonas paucimobilis; WBC: White
blood cell; ISPD GL: International Society for Peritoneal Dialysis Guidelines; UV:
Ultraviolet; EM: Electron microscopy.

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Authors’ contributions
CK, KM, YK, TH and AO participated in discussions of the patient’s case. CK
drafted and is responsible for the final version of the manuscript. All authors
read and approved the manuscript and agree with its submission to this
journal. All authors read and approved the final manuscript.

Availability of data and materials
All data and materials were included in the manuscript.

Declarations

Ethics approval and consent to participate
This report was written in compliance with the Declaration of Helsinki. For this
type of case report, ethics approval is not required.

Consent for publication
Written informed consent was obtained from the patient’s family for the
publication of this case report.

Competing interests
The authors declare that they have no competing interests.
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