A Case of Intra-abdominal abscess due to Sphingomonas paucimobilis in a patient on Peritoneal dialysis: A case report and review of literature

Abstract

*Sphingomonas paucimobilis* is an aerobic gram-negative bacillus, widely distributed in the water and soil. It has also been found in nosocomial environments causing nosocomial infections. *S. paucimobilis* is a rare cause of peritoneal dialysis (PD)-related peritonitis. Here, we report the 14th case, with a literature review. Our case is unique as this is the first reported case of intra-abdominal abscess associated with *S. paucimobilis* PD-related peritonitis.

Keywords: End-stage renal disease, peritoneal dialysis, peritonitis, *Sphingomonas paucimobilis*

Introduction

We present the first reported cause of intra-abdominal abscess associated with *Sphingomonas paucimobilis* PD-related peritonitis. *Sphingomonas paucimobilis* PD-related peritonitis is also rare with only 13 cases reported in the medical literature before this.

Case History

A 35-year-old male patient on peritoneal dialysis (PD) presented in January 2019 with a 1-day history of abdominal pain and cloudy PD dialysate. He had end-stage kidney disease from chronic glomerulonephritis and was started on automated PD in May 2015. Other medical history included hypertension and a history of inguinal herniorrhaphy. There was an episode of culture-negative peritonitis in 2016. At the time of presentation, his PD regimens consisted of 6 cycles at night, with a fill volume of 2.1 L and last fill of 1.2 L using icodextrin. His daily ultrafiltration volume was around 1.5 L. Two weeks prior to the current presentation, he had an episode of culture-negative peritonitis, which resolved with 2 weeks of intraperitoneal (IP) ampicillin/sulbactam and ceftazidime. On examination, he was afebrile and blood pressure was 110/70 mmHg. There was periumbilical tenderness but no guarding. Other systemic examination was unremarkable. His PD catheter exit site was normal. PD effluent was turbid, and its white blood cells (WBCs) was 640/µL with 100% neutrophils. Gram stain of the fluid did not show any organism. Empirical IP ampicillin/sulbactam and ceftazidime were started. Three days later, the PD fluid WBC count came down to 350 with 91% neutrophils. At the same time, the PD fluid culture grew *Sphingomonas paucimobilis*, sensitive only to ciprofloxacin, netilmicin, and co-trimoxazole; but resistant to gentamicin, amikacin, ceftazidime, piperacillin, imipenem, and meropenem. In view of clinical stability, improvement in PD fluid WBC count and the uncertain clinical significance of the organism, the patient was continued on IP ampicillin/sulbactam and ceftazidime.

By day 7, the patient still had abdominal pain and cloudy effluent. Repeat PD fluid was still positive for *S. paucimobilis*, thus antibiotic was changed to intravenous (IV) ciprofloxacin 200 mg BD. Within 72 h, the PD fluid became clear and WBC count became 0. Despite this, the PD fluid remained positive for *S. paucimobilis*, possibly because of the presence of biofilm on the catheter. Ultrasound tunnel tract did not reveal any collection. In view of persistent abdominal pain, an ultrasound of the abdomen was done which showed a 13.2 by 10.6 by 10.8 cm loculated abscess around the catheter tip in the pelvis. IV netilmicin 150 mg OD was then added, and the patient underwent PD catheter removal.
and drainage of the abscess on day 16. Following this, there was marked clinical improvement with resolution of abdominal pain and down trending of his inflammatory markers. He was shifted to hemodialysis via a right internal jugular tunneled dialysis catheter and was discharged well with continuation of IV ciprofloxacin and netilmicin for a total duration of 35 and 19 days, respectively. After completion of the antibiotics, a follow-up ultrasound scan showed total resolution of the intra-abdominal abscess.

Discussion

*S. paucimobilis* is a yellow-pigmented, aerobically gram-negative bacillus that has a single polar flagellum with slow motility.[1] First isolated in a human infection and named *Pseudomonas paucimobilis* in 1977,[3] it was then reclassified and renamed as *S. paucimobilis* in 1990.[3] There are currently more than 30 species in the genus *Sphingomonas*, with *S. paucimobilis* considered as the main pathogenic species. Widely distributed in the natural environments in water and soil, the organisms have been isolated in multiple environments—sea ice, river water, mineral water, and other water distribution systems. They are also found in nosocomial environments, such as laboratory instruments, ventilators, and hospital water systems.[4]

Human infections caused by *S. paucimobilis* are generally rare. *S. paucimobilis* can cause both sporadic and nosocomial infections. Sporadic infections include infected leg ulcers, urinary tract infection, brain and spleen abscesses. Nosocomial infections include bacteremia caused by contaminated solutions, for example, distilled water, hemodialysis fluid, and sterile drug solutions.[5] Most of the infections are in immunocompromised patients with indwelling devices. No deaths due to infections from this organism have been reported so far. It is a unique organism with low virulence thought to be secondary to the lack of lipid A in its outer membrane; instead there is an atypical lipopolysaccharide sphingoglycolipid.[6] The absence of these components may therefore explain the favorable prognosis observed in the previously reported cases.

Although nosocomial infection with *S. paucimobilis* has been infrequently reported, PD-related peritonitis with this organism is exceptionally rare. Only 13 cases of *S. paucimobilis* PD-related peritonitis have been reported prior to ours.[7-18] Table 1 shows the clinical features of

| Reference              | Country       | Age (month) | PD vintage (month) | Previous peritonitis | Presenting complaints                                      | Temperature at presentation (°C) | Etiology of ESRD                        |
|------------------------|---------------|-------------|--------------------|----------------------|-----------------------------------------------------------|----------------------------------|----------------------------------------|
| Glupczynski et al., 1984[1] | Belgium      | 74          | 6                  | NR                   | AP, CD, vomiting                                           | 36.6                             | Analgesic nephropathy                  |
| Glupczynski et al., 1984[1] | Belgium      | 33          | 13                 | Yes                  | AP, CD                                                   | NR                               | Chronic pyelonephritis                |
| Swann et al., 1985[9]    | United Kingdom| 61          | 33                 | Yes                  | CD                                                        | NR                               | Hypertensive nephrosclerosis           |
| Baddour et al., 1985[9]  | USA           | 50          | 36                 | No                   | CD                                                       | NR                               | NR                                     |
| Nguyen et al., 1987[10]  | USA           | 65          | 28                 | Yes                  | AP, CD, nausea                                            | NR                               | Glomerulo-nephritis                   |
| De Paoli Vitali et al., 1988[11] | Italy     | 38          | 36                 | Yes                  | AP, CD, nausea                                            | NR                               | Unknown                               |
| Phillips et al., 1990[12] | United Kingdom| 64          | 36                 | Yes                  | CD                                                       | NR                               | Hypertensive nephrosclerosis           |
| Dervisoglu et al., 2008[13] | Turkey       | 50          | 60                 | Yes                  | AP, CD                                                   | NR                               | NR                                     |
| Tambawala et al., 2011[14] | Pakistan     | 3.5         | 15                 | NR                   | CD, decreased oral intake, oral ulcers, perineal rash     | NR                               | Prune Belly syndrome                  |
| Lee et al., 2013[15]     | Korea         | 63          | 72                 | Yes                  | AP, CD                                                   | 36.8                             | Diabetic nephropathy, hypertensive nephrosclerosis |
| Mohan et al., 2015[16]   | United Arab Emirates | 50    | 28                 | Yes                  | AP, CD, fever, vomiting                                   | 36.8                             | NR                                     |
| Owen et al., 2016[17]    | USA           | 35          | 24                 | NR                   | AP, CD                                                   | NR                               | Collapsing focal segmental glomerulo-sclerosis |
| Yilmaz et al., 2018[18]  | Turkey        | 63          | 41                 | No                   | AP, CD, fever, vomiting                                   | 37.2                             | Hypertensive nephrosclerosis           |
| Present case             | Brunei        | 35          | 43                 | Yes                  | AP, CD                                                   | NR                               | Chronic glomerulo-nephritis           |

ESRD: End-stage renal disease; NR: Not reported; AP: Abdominal pain; CD: Cloudy dialysate
these patients. Vintage on PD ranged from 6 to 72 months. Almost all patients presented with abdominal pain and cloudy dialysate. Only two (14%) patients reported having fever on presentation. Half of them had a recent episode of peritonitis, ranging from 1 week to 7 months before their episodes caused by S. paucimobilis. Table 2 shows their laboratory findings, which vary greatly among the 14 patients. Our case is unique as this is the first reported case of intra-abdominal abscess associated with S. paucimobilis peritonitis. The treatment and outcomes have also been variable as shown in Table 3. Half of the cases were cured with appropriate antibiotics, but the other half required catheter removal, ranging from day 7 to 21 from the day of diagnosis, to eradicate the infection. Most of the latter patients had clinically improved with antibiotics but continued to have positive cultures in their dialysates. This is an exceptionally high treatment failure rate despite the organism’s low virulence. This high therapeutic failure rate is partly due to the organism’s unpredictable antibiotic sensitivity. None of the case reports have shown any consistent pattern of antibiotic sensitivity. Hence to this date, no definitive guidelines exist for antimicrobial therapy for S. paucimobilis infections. Trimethoprim-sulfamethoxazole, chloramphenicol, ciprofloxacin, and aminoglycosides have all been successfully used to eradicate this bacterium. Among the seven cases that were cured, five used an IP aminoglycoside. Although an aminoglycoside plus a third-generation cephalosporin or a carbapenem alone could effectively treat S. paucimobilis infections,[19] there is insufficient data to comment on the use of monotherapy versus combination therapy. International Society of Peritoneal Dialysis guidelines on the management of peritonitis recommend removing the catheter if there is no clinical improvement within 5 days of appropriate antibiotic therapy. This same criterion should be applied to S. paucimobilis peritonitis.

Our case is unique as firstly the organism was resistant to carbapenem and gentamicin which were both successfully used in previous cases. Secondly, this is the first reported case of intra-abdominal abscess associated with S. paucimobilis PD-related peritonitis. Due to the presence of this intra-abdominal abscess as well as persistent abdominal pain and failure to clear the organism from the effluent, he required PD catheter removal to eradicate the infection.

In conclusion, we report the first case of intra-abdominal abscess associated with S. paucimobilis PD-related peritonitis, which was treated by catheter removal. Despite having low virulence, treatment failure rate of this peritonitis is extremely high, with removal of the catheter in 50% of reported cases. This case adds to the current literature of peritonitis caused by S. paucimobilis.

### Table 2: Laboratory Findings in 14 Patients with Sphingomonas paucimobilis PD-Related Peritonitis

| Reference | WBC (per µL) | Initial Gram stain | Culture (day at which it turns positive) | WBC (>10³/L) | CRP (mg/dL) | ESR (mm/h) |
|-----------|--------------|-------------------|----------------------------------------|--------------|-------------|------------|
| Glupczynski et al., 1984[7] | 850 | Gram negative bacilli | P. paucimobilis (NR) | NR | NR | NR |
| Glupczynski et al., 1984[7] | 9100 | Gram negative bacilli | P. paucimobilis (NR) | NR | NR | NR |
| Swann et al., 1985[9] | NR | Gram negative rods | P. paucimobilis (D2) | NR | NR | NR |
| Baddour et al., 1985[9] | 130 (NE 82%) | NR | P. paucimobilis (NR) | NR | NR | NR |
| 1st relapse: 550 (NE 92%) | 1st relapse: NR | 1st relapse: negative | | | | |
| 2nd relapse: NR | 2nd relapse: NR | 2nd relapse: P. paucimobilis (NR) | NR | NR | NR | NR |
| Nguyen et al., 1987[10] | NR | NR | P. paucimobilis (NR) | NR | NR | NR |
| De Paoli Vitali et al., 1988[11] | 4000 | NR | P. paucimobilis (NR) | NR | NR | NR |
| Phillips et al., 1990[12] | 152 (NE 75%) | Gram negative bacilli | P. paucimobilis (D2) | NR | NR | NR |
| Dervisoglu et al., 2008[13] | 148 (NE 90%) | No organism | S. paucimobilis (D3) | 8.5 | 9.35 | 82 |
| Tambwala et al., 2011[14] | 600 (NE 80%) | Gram negative rods | S. paucimobilis (D1) | 7.7 | NR | NR |
| Lee et al., 2013[15] | 2040 (NE 85%) | No organism | S. paucimobilis (NR) | 7.6 | 5.9 | NR |
| 1st relapse: 2880 (NE 75%) | 1st relapse: NR | 1st relapse: S. paucimobilis (NR) | | | | |
| Mohan et al., 2015[16] | NR (NE 80%) | Gram negative rods | S. paucimobilis (D4) | 15.4 | NR | NR |
| Owen et al., 2016[17] | 2835 (NE 47%) | NR | S. paucimobilis (D3) | NR | NR | NR |
| 1st relapse: 87 (NE 4%) | 1st relapse: S. paucimobilis (NR) | | | | | |
| Yılmaz et al., 2018[18] | 4350 (NE 55%) | No organism | S. paucimobilis (D3) | NR | NR | NR |
| Present case | 640 (NE 100%) | No organism | S. paucimobilis (D3) | 7.4 | 6 | NR |

P. paucimobilis: Pseudomonas paucimobilis; NR: Not reported; NE : % of neutrophil; S. paucimobilis: Sphingomonas paucimobilis.
| Reference                  | Initial empirical treatment (dose)                                                                 | Maintenance treatment (dose)                                                                 | Total days of treatment | Outcome                                      |
|---------------------------|---------------------------------------------------------------------------------------------------|---------------------------------------------------------------------------------------------|-------------------------|-----------------------------------------------|
| Glupczynski et al., 1984[7] | NR                                                                                               | TMP-SMX IP (16 mg/L and 80 mg/L respectively)                                                | 14                      | Dialysate clear by day 2 Resolved             |
| Glupczynski et al., 1984[7] | Cefazolin IP (50 mg/L) + Tobramycin IP (NR)                                                       | 1) Ampicillin IP (50 mg/L)                                                                  | 1) 5                    | Dialysate clear by day 5                     |
| Swann et al., 1985[8]     | Vancomycin IP (12.5 mg/L) + Gentamicin IP (4 mg/L)                                                | 2) Amoxicillin PO (3g/day)                                                                  | 2) 7                    | Catheter removed on day 12                   |
| Baddour et al., 1985[9]   | Cephalothin IP (1 g)                                                                              | 3) After catheter removal, tobramycin IV (80 mg)                                             | 3) NR                   | Back on PD later                             |
| Nguyen et al., 1987[10]   | Vancomycin IP (15 mg/kg) + Tobramycin IP (1.75 mg/kg) + Ampicillin (route NR)                   | 1) Mezlocillin + cefoxitin (route NR)                                                        | 1) 13                   | Catheter removed (day NR)                    |
| De Paoli Vitali et al., 1988[11] | Cephalothin IP (1 g/exchange loading) + Tobramycin IP (1.7 mg/kg loading)                     | Cephalothin IP (250 mg/L) + Tobramycin IP (8 mg/L)                                         | NR                      | Dialysate clear by day 3 but cloudy again on day 7 |
| Phillips et al., 1990[12]  | Ciprofloxacin PO (250mg TDS)                                                                    | Netilmicin IP (10 mg/L 4 times a day)                                                        | NR                      | Resolved                                      |
| Dervisoglu et al., 2008[13] | Vancomycin IP (2g single dose)                                                                 | 1) Imipenem IV (500 mg/day) + Gentamicin IP (80 mg loading then 40 mg/day)                | 1) 18                   | Dialysate clear by day 2 but persistent *S. paucimobilis* in PD fluid on day 5, 7, 10 and 17 (despite dialysate clear and without WBCs) |
| Tambawala et al., 2011[14] | Amikacin IP (2 mg/kg/bag)                                                                       | 2) After catheter removal, imipenem IV                                                      | 2) 7                    | Catheter removed on day 21                   |
| Lee et al., 2013[15]      | Cefazolin IP (1 g/day) + Ceftazidime IP (1 g/day)                                               | 1) Amikacin IP (25 mg/L)                                                                    | 1) 4                    | Resolved                                      |
| Mohan et al., 2015[16]    | Vancomycin IP (1 g single dose) + Ciprofloxacin IV (NR)                                         | 2) Meropenem IV (NR)                                                                        | 2) 7                    | Cloudy dialysate again on day 15              |
| Owen et al., 2016[17]     | Vancomycin IP (NR) + Ceftazidime IP (NR)                                                         | 1st relapse: Ciprofloxacin PO (NR) + Ceftriaxone IP (NR)                                    | 1st relapse:10          | Catheter removed on day 17                   |
| Yilmaz et al., 2018[18]   | Vancomycin IP (1 g single dose) + Ceftazidime IP (1 g/day)                                      | 1) Ciprofloxacin IV (200 mg BD)                                                             | 1) 35                   | Dialysate clear by day 5 Resolved             |
| Present case              | Ampicillin/sulbactam IP (1.5 g BD) + Ceftazidime IP (1 g/day)                                   | 2) Netilmicin IV (150mg OD)                                                                 | 2) 19                   | Catheter removed on day 16                   |

NR: Not reported; TMP-SMX: Trimethoprim-sulfamethoxazole; IP: Intraperitoneally; PO: Orally; IV: Intravenously
Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Winn WC, Allen SD, Janda WM, Koneman EW, Procop GW, Schreckenberger PC, et al. Koneman’s Color Atlas and Textbook of Diagnostic Microbiology. 6th ed. Philadelphia: Lippincott Williams and Wilkins; 2006.
2. Holmes B, Owen RJ, Evans A, Malnick H, Willcox WR. Pseudomonas paucimobilis, a new species isolated from human clinical specimens, the hospital environment, and other sources. Int J Syst Bacteriol 1977;27:133-46.
3. Yabuuchi E, Yano I, Oyaizu H, Hashimoto Y, Ezaki T, Yamamoto H. Proposals of Sphingomonas paucimobilis gen. nov. and comb. nov., Sphingomonas parapaucimobilis sp. nov., Sphingomonas yanoikuyae sp. nov., Sphingomonas adhaesiva sp. nov., Sphingomonas capsulata comb. nov., and two genospecies of the genus Sphingomonas. Microbiol Immunol 1990;34:99-119.
4. Pascale R, Russo E, Esposito I, Leone S, Esposito S. Sphingomonas paucimobilis osteomyelitis in an immunocompetent patient. A rare case report and literature review. New Microbiol 2013;36:423-6.
5. Reina J, Bassa A, Llompart I, Portela D, Borrell N. Infections with Pseudomonas paucimobilis: Report of four cases and review. Rev Infect Dis 1991;13:1072-6.
6. Ryan MP, Adley CC. Sphingomonas paucimobilis: a persistent Gram-negative nosocomial infectious organism. J Hosp Infect 2010;75:153-7.
7. Głupczyński Y, Hansen W, Dratwa M, Tielemans C, Wens R, Collart F, et al. Pseudomonas paucimobilis peritonitis in patients treated by peritoneal dialysis. J Clin Microbiol 1984;20:1225-6.
8. Swann RA, Foulkes SJ, Holmes B, Young JB, Mitchell RG, Reeder ST. Agrobacterium yellow group and Pseudomonas paucimobilis causing peritonitis in patients receiving continuous ambulatory peritoneal dialysis. J Clin Pathol 1985;38:1293-9.
9. Baddour LM, Kraus AP Jr, Smalley DL. Peritonitis due to Pseudomonas paucimobilis during ambulatory peritoneal dialysis. South Med J 1985;78:366.
10. Nguyen V, Swartz RD, Reynolds J, Wilson D, Port FK. Successful treatment of Pseudomonas peritonitis during continuous ambulatory peritoneal dialysis. Am J Nephrol 1987;7:38-43.
11. De Paoli Vitali E, Rossi MR, Farinelli A. Pseudomonas-like species I1K-1 peritonitis in peritoneal dialysis. Nephron 1988;48:337.
12. Phillips G, Fleming LW, Stewart WK. Pseudomonas paucimobilis peritonitis in a patient on CAPD successfully treated with ciprofloxacin and netilmicin. Eur J Clin Microbiol Infect Dis 1990;9:630-1.
13. Derivisoglu E, Meric M, Kalender B, Sengul E. Sphingomonas paucimobilis peritonitis: A case report and literature review. Perit Dial Int 2008;28:547-50.
14. Tambawala AQ, Hamid S, Khan I, Ali A. Continuous ambulatory peritoneal dialysis (CAPD) associated peritonitis in a child: A rare case of peritonitis caused by sphingomonas paucimobilis. J Pak Med Assoc 2011;61:178-80.
15. Lee JU, Kim JK, Yun SH, Park MS, Lee NE, Sun JO, et al. A case of peritoneal dialysis-associated peritonitis caused by Sphingomonas paucimobilis. Kidney Res Clin Pract 2013;32:78-80.
16. Mohan D, Railey M. Sphingomonas paucimobilis peritonitis: A case report and review of the literature. Saudi J Kidney Dis Transpl 2015;26:567-71.
17. Owen J, Washco V, Reisin E. Successful return to peritoneal dialysis after a case of relapsing Sphingomonas paucimobilis peritonitis. Clin Nephrol 2016;86:287-89.
18. Yilmaz F, Bora F, Eroş F. Peritoneal dialysis related peritonitis by Sphingomonas Paucimobilis. Ther Apher Dial 2018;22:205-6.
19. Hsueh PR, Teng LJ, Yang PC, Chen YC, Pan HJ, Ho SW, et al. Nosocomial infections caused by Sphingomonas paucimobilis: Clinical features and microbiological characteristics. Clin Infect Dis 1998;26:676-81.