Scientific Research Publishing

Open Journal of Nephrology, 2022, 12, 228-234
https://www.scirp.org/journal/ojneph
ISSN Online: 2164-2869
ISSN Print: 2164-2842

Successful Treatment of Peritoneal Dialysis Related Peritonitis from Multi-Drug Resistant *Sphingomonas paucimobilis* with Combination Therapy: A Case Report

Gennaro Argentino[1*], Silvio Borrelli[2], Ciro Paglionico[1], Andrea Camocardi[1], Mario Iorio[1], Alessandra Antonia Mele[1], Andrea Pota[1], Adelia Sagliocca[1], Stefania Brancaccio[1], Lucia Di Micco[1]

[1]"Ospedale del Mare", Nephrology and Dialysis Unit, ASL Napoli 1 Centro, Naples, Italy
[2]Nephrology and Dialysis Unit, University of Studies of Campania "Luigi Vanvitelli", Naples, Italy
Email: *gennaro_ag@libero.it

How to cite this paper: Argentino, G., Borrelli, S., Paglionico, C., Camocardi, A., Iorio, M., Mele, A.A., Pota, A., Sagliocca, A., Brancaccio, S. and Di Micco, L. (2022) Successful Treatment of Peritoneal Dialysis Related Peritonitis from Multi-Drug Resistant *Sphingomonas paucimobilis* with Combination Therapy: A Case Report. *Open Journal of Nephrology*, **12**, 228-234.
https://doi.org/10.4236/ojneph.2022.122023

Received: May 29, 2022
Accepted: June 27, 2022
Published: June 30, 2022

Abstract

Sphingomonas paucimobilis is an emerging gram-negative aerobic bacterium, generally causing infections in immunocompromised patients. Few data are available about peritonitis in peritoneal dialysis due to this pathogen. The clinical courses and outcomes of peritonitis are variable, with a high frequency of catheter removal and peritoneal dialysis withdrawal. No guidelines are available for the treatment of *Sphingomonas paucimobilis* related peritonitis, due to its emerging role as pathogen, the high antibiotic resistance and unpredictable antibiotic sensitivity. Here, we describe a case of *Sphingomonas paucimobilis* peritonitis in a 52-year-old diabetic patient in Continuous Cycler-Assisted Peritoneal Dialysis (CCPD) for 4 months, successfully treated with a combined intraperitoneally administration of meropenem (250 mg/L) and ciprofloxacin (100 mg/L) for 21 days. No hospital admission and change of peritoneal dialysis scheme were needed; no relapses of peritonitis were observed during 18 months of follow-up.

Keywords

Sphingomonas paucimobilis, Peritoneal Dialysis, Peritonitis, Antibiotic Therapy

1. Introduction

Sphingomonas paucimobilis is an aerobic, non-spore-forming gram-negative bacillus with a single polar flagellum and slow motility. Initially classified as *Pseu-*

domonas paucimobilis in 1977, it was subsequently reclassified as *Sphingomonas* on the basis of phylogenetic data [1] [2].

Sphingomonas paucimobilis is widely distributed in water and soil and rarely reported in clinical setting [3]. It is thought to be an opportunistic pathogen and has been associated to bacteremia and septicemia caused by contaminated solutions, e.g. distilled water, dialysis solutions, other than infected ulcers in the lower limbs, urinary tract infection, brain and splenic abscesses.

To the best of our knowledge, there are few data regarding *Sphingomonas paucimobilis* infections in Peritoneal Dialysis (PD) patients, making difficult the treatment of peritonitis caused by this pathogen.

We report a case of successful treatment of *Sphingomonas paucimobilis* PD related peritonitis.

2. Case Report

A 52-years-old male PD patient presented to our outpatient clinic for a 1-day history of fever and cloudy peritoneal dialysate drainage. He had end stage renal disease due to diabetic nephropathy and had been receiving CCPD scheme for four months (three glucose-bags for a total amount of thirteen litres at night and 1.5-litres bag daily of icodextrin) and no previous history of peritonitis.

On physical examination, his abdomen was painful, with no vomiting or intestinal occlusion signs, and temperature was 38˚C. His peritoneal catheter and the exit site were normal. Laboratory blood tests showed leukocytes 13,410/mm^3 (90.7% Neutrophils), C-Reactive Protein (CRP) 6.3 mg/dL and Procalcitonin 0.77 ng/mL (see **Table 1**). Peritoneal fluid analysis revealed numerous white blood cells (4370 mmc), mainly neutrophils (80%). The peritoneal effluent was tested for bacterial culture in "BACTEC" (**Figure 1**). The patient was not admitted to our hospital and we started on empiric therapy for peritonitis with 250 mg of piperacillin-tazobactam per liter of dialyser solution and 20 mg of teicoplanin per liter of dialyser solution, both intraperitoneally. The peritoneal effluent bacterial culture was positive after four days of incubation, and the subcultures, once inoculated to solid media, became positive for a gram negative, non-fermenting, and catalase- and oxidase-positive bacteria. The bacterium detected was *Sphingomonas paucimobilis*. The susceptibility testing showed antibiotic sensitivity to cefepime, ceftazidime, levofloxacin and meropenem and resistance to amikacina and imipenem.

Table 1. Laboratory tests.

	Day-1	Day-3	Day-6
Leukocytes	13,410/mm^3 (90.7% Neutrophils)	6.9×10^3/mm^3	6.95×10^3/mm^3
CRP	6.3 mg/dL	2.04 mg/dL	CRP 0.06 mg/dL
Procalcitonin	0.77 ng/mL	0.97 ng/mL	0.25 ng/mL
Peritoneal fluid analysis white blood cells	4370/mmc (80% neutrophils)	1302.0/mmc (80% neutrophils)	15.0/mmc

Sample: **PERITONEAL LIQUID; AEROBES**
Outcome: Development of microorganisms

Biochemical Identification		
Isolated microorganism:	**Sphingomonas paucimobilis**	
ANTIBIOGRAM reported according to EUCAST interpretative criteria.		
Antibiotic	**MIC**	**SRI**
Amikacin		**(R)**
Cefepime		**(S)**
Ceftazidime		**(S)**
Imipenem		**(R)**
Meropenem		**(S)**
Piperacillin/tazobactam		**(I)**
trimethoprim sulfamethoxazole		**(R)**
Levofloxacin		**(S)**

Legend: **S** Sensitive; **I** Intermediate; **R** Resistant; **IE** Insufficient evidence that the species is a good target for therapy. MICs can be reported without interpretation.

Figure 1. Culture test.

After three days from the beginning of the empiric antibiotic therapy, the patient presented an improvement in the clinical symptoms, with the disappearance of abdominal pain, resolution of fever and neutrophilic leukocytosis. Leukocytes were still present in the dialysate drainage (even at the fifth day of observation), with CRP and procalcitonin levels persistently high and a decrease in peritoneal ultrafiltrate (see **Table 1**).

According to the susceptibility testing, we discontinued piperacillin-tazobactam and teicoplanin and started an intraperitoneal therapy with meropenem (250 mg/L) and ciproxin (100 mg/L), and the patient continued performing his usual CCPD scheme.

After three days of treatment, the microscopic analysis of the peritoneal fluid showed less of five 15 leucocytes/mmc, without bacterial growth, in association with a significant decrease of serum CRP and normalization of serum procalcitonin (see **Table 1**). The antibiotic therapy was maintained for up to three weeks. The subsequent analyses showed sterile dialysate drainage on day 7 and on day 14 after the end of the antibiotic treatment with intraperitoneal meropenem and ciprofloxacin. No relapse of peritonitis occurred during 18 months of follow-up.

3. Discussion

This case report shows successful treatment of peritonitis by *Sphingomonas paucimobilis* by intraperitoneal administration of meropenem and ciprofloxacin in a

peritoneal patient on a CCPD scheme, not hospitalized.

Sphingomonas paucimobilis is widely distributed in nature, thus in soil and aquatic environments, such as drinking and distilled water [4]. It has been also found in nosocomial environments, but infrequently in hemodialysis fluids. Nevertheless, it rarely affects humans but it is associated with high antibiotic resistance [5]. Moreover, *Sphingomonas* are able to form dense biofilms with the risks of relapsing and/or being resistant to catheter-associated infections [6].

Currently, no recommendation is available for antimicrobial therapy against *Sphingomonas* infections probably because it is an emerging pathogen with an unpredictable antibiotic sensitivity and a high antibiotic resistance. Additionally, PD-related peritonitis caused by this organism is very rare [7].

To our knowledge, there are only 14 cases reporting *Sphingomonas paucimobilis* associated peritonitis. As shown in **Table 2** [8]-[19], clinical outcomes in infected patients were heterogeneous: indeed, a complete resolution of the infection was reported in 8 patients, whereas in the remaining 6 patients, peritoneal catheter was removed and PD was discontinued.

In the case of success of antibiotic therapy, the intravenous route was used in 1 out of 8 cases, whereas the intraperitoneal route was used in 7 cases.

The antibiotic classes used in this infection were heterogeneous, including trimethoprim plus sulfamethoxazole, carbapenems, ciprofloxacin, and aminoglycosides.

Considering the lack of uniformity in the class of antibiotics and route of administration, as well as the antibiogram (aminoglycosides and imipenem resistance), we decided to use a combination of meropenem and ciprofloxacin by intraperitoneal route for a duration of three weeks. This antibiotic schedule allowed us to maintain our patient in CCPD with no need of hospital admission. Furthermore, no relapses were reported after 18 months of follow-up.

As reported in other cases (**Table 2**), we were not able to identify the source of infection, because the pathogen was isolated neither in the dialysis solution, nor in other solutions used for our patients. Considering that the patient started dialysis after only four months, we cannot exclude that infection was contracted during surgery for peritoneal catheter, though no cases were reported in our hospital [20].

In conclusion, *Sphingomonas paucimobilis* is a virulent and multi-drug resistant pathogen that may seldom induce a harmful peritonitis in PD patients, leading to treatment failure and catheter removal in the majority of patients [21]. In our case, the intraperitoneal administration of an association of meropenem and ciprofloxacin was able to eradicate peritonitis, with no need of hospitalization, and allowed us to maintain our patient on his CCPD scheme.

No definitive guidelines are available to treat PD-related peritonitis caused by *Sphingomonas paucimobilis*. Various antimicrobials (single or associations) and routes of administration were described, leading to different outcomes. The high antibiotic resistance and the inadequate dosage of antibiotics (to avoid an impairment of residual renal function) might be responsible for the high rate of

Table 2. Summary of reported peritoneal dialysis-related peritonitis cases caused by *Sphingomonas paucimobilis* spp. Legend: IP: intraperitoneally; IV: intravenously; NR: not revealed.

Case no	References	Year	Gender	Age	Catheter removal	Antimicrobials	Route of Administration	Outcome
1	(8)	1984	Female	74	No	Trimethoprim Sulfamethoxazole Ampicillin	**IP**	Cured
2	(8)	1984	Male	33	Yes	1) Cefazolin IP + tobramycin IP (duration NR) 2) Ampicillin IP for 5 days 3) Amoxicillin orally for 7 days 4) After catheter removal, tobramycin IV (duration NR)	**IP, IV**	Catheter removed on day 12 after the start of antibiotic therapy
3	(9)	1985	Male	61	No	Vancomycin IP + gentamicin IP (duration NR)	**IP**	Cured
4	(10)	1985	Male	50	No	1) Cephalothin IP for 4 days 2) Tobramycin IP for 14 days	**IP**	Cured
5	(10)	1987	Female	65	Yes	1) Vancomycin for 10 days + tobramycin 12 days + ampicillin for 3 days (route NR) 2) Mezlocillin for 13 days + cefoxitin for 13 days (route NR) 3) Chloramphenicol for 13 days (route NR)	**IP**	Catheter removed (day NR)
6	(11)	1988	Female	38	Yes	Tobramycin Ciprofloxacin	**IP**	Catheter removed on 7 day
7	(12)	1990	Male	64	No	1) Ciprofloxacin orally for 3 days 2) Netilmicin IP (duration NR)	**OS, IP**	Cured
8	(13)	2008	Male	50	Yes	1) Vancomycin IP, single dose 2) Imipenem IV + gentamicin IP for 18 days 3) After catheter removal, imipenem IV for 7 days	**IP, IV**	Catheter removed on day 21 after the start of antibiotic therapy
9	(14)	2011	Female	3.5	No	1) Amikacin IP for 4 days 2) Meropenem IV for 7 days	**IP, IV**	Cured
10	(15)	2013	Male	63	Yes	Cefazolin IP (1 g/day) + Ceftazidime IP (1 g/day) 1st relapse: Imipenem IP (1 g)	**IP**	Catheter Removed Hemodialysis
11	(16)	2015	Female	50	No	1) Tobramycin for 21 days 2) Meropenem IV for 21 days	**IV**	Cured
12	(17)	2016	Female	50	Yes	Ceftazidime	**IP**	Cured
13	(18)	2018	Female	63	No	Ceftazidime Amikacin	**IP**	Cured

Continued

14	(19)	2019	Male	35	Yes	intraperitoneal (IP) ampicillin/sulbactam and ceftazidime	**IP**	Catheter Removed Hemodialysis
15	Our study	2019	Male	52	No	Ciprofloxacin Meropenem	**IP**	Cured

treatment failure with this new-emerging organism. Combination therapy of antimicrobials might be beneficial to overcome antibiotic resistance, but more studies are needed to confirm our success and develop treatment guidelines.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

References

[1] Winn, W.C., Allen, S.D., Janda, W.M., *et al.* (2006) Koneman's Color Atlas and Textbook of Diagnostic Microbiology. 6th Edition, Lippincott Williams & Wilkins, Philadelphia.

[2] Holmes, B., Owen, R.J., Evans, A., Malnick, H. and Willcox, W.R. (1977) *Pseudomonas paucimobilis*, a New Species Isolated from Human Clinical Specimens, the Hospital Environment, and Other Sourced. *International Journal of Systematic Bacteriology*, **27**, 33-46. https://doi.org/10.1099/00207713-27-2-133

[3] Pascale, R., Russo, E., Esposito, I., Leone, S. and Esposito, S. (2013) *Sphingomonas paucimobilis* Osteomyelitis in an Immunocompetent Patient: A Rare Case Report and Literature Review. *New Microbiologica*, **36**, 423-426.

[4] Bayram, N., Devrim, I., Apa, H., Gülfdan, G., Türkyılmaz, H.N. and Günay, I. (2013) *Sphingomonas paucimobilis* Infections in Children: 24 Case Reports. *Mediterranean Journal of Hematology and Infectious Diseases*, **5**, e2013040. https://doi.org/10.4084/mjhid.2013.040

[5] Lin, J.N., Lai, C.H., Chen, Y.H., Lin, H.L., Huang, C.K., Chen, W.F., Wang, J.L., Chung, H.C., Liang, S.H. and Lin, H.H. (2010) *Sphingomonas paucimobilis* Bacteremia in Humans: 16 Case Reports and a Literature Review. *Journal of Microbiology, Immunology and Infection*, **43**, 35-42. https://doi.org/10.1016/S1684-1182(10)60005-9

[6] Lee, J.S., Shin, Y.K., Yoon, J.H., Takeuchi, M., Pyun, Y.R. and Park, Y.H. (2001) *Sphingomonas aquatilis* sp. nov., *Sphingomonas koreensis* sp. nov. and *Sphingomonas taejonensis* sp. nov., Yellow-Pigmented Bacteria Isolated from Natural Mineral Water. *International Journal of Systematic and Evolutionary Microbiology*, **51**, 1491-1498. https://doi.org/10.1099/00207713-51-4-1491

[7] Yılmaz, F., Bora, F. and Ersoy, F. (2018) Peritoneal Dialysis Related Peritonitis by *Sphingomonas paucimobilis*. *Therapeutic Apheresis and Dialysis*, **22**, 205-206. https://doi.org/10.1111/1744-9987.12637

[8] Kawasaki, S., Moriguchi, R., Sekya, K., *et al.* (1994) The Cell Envelope Structure of the Lipopolysaccharide-Lacking Gram-Negative Bacterium *Sphingomonas paucimobilis*. *Journal of Bacteriology*, **176**, 284-290. https://doi.org/10.1128/jb.176.2.284-290.1994

[9] Glupczynski, Y., Hansen, W., Dratwa, M., *et al.* (1984) *Pseudomonas paucimobilis* Peritonitis in Patients Treated by Peritoneal Dialysis. *Journal of Clinical Microbiology*, **20**, 1225-1226. https://doi.org/10.1128/jcm.20.6.1225-1226.1984

[10] Swann, R.A., Foulkes, S.J., Holmes, B., Young, J.B., Mitchell, R.G. and Reeders, S.T. (1985) Agrobacterium Yellow Group and *Pseudomonas paucimobilis* Causing Peritonitis in Patients Receiving Continuous Ambulatory Peritoneal Dialysis. *Journal of Clinical Pathology*, **38**, 1293-1299. https://doi.org/10.1136/jcp.38.11.1293

[11] Tambawala, A.Q., Hamid, S., Khan, I. and Ali, A. (2011) CAPD Associated Peritonitis in a Child: A Rare Case of Peritonitis Caused by *Sphingomonas paucimobilis*. *Journal of Pakistan Medical Association*, **61**, 178-180.

[12] Baddour, L.M., Kraus, A.P. and Smalley, D.L. (1985) Peritonitiis Due to *Pseudomonas paucimobilis* during Ambulatory Peritoneal Dialysis. *Southern Medical Journal*, **78**, 366. https://doi.org/10.1097/00007611-198503000-00044

[13] Reina, J., Bassa, A., Llompart, I., Portela, D. and Borrell, N. (1991) Infections with *Pseudomonas paucimobilis*: Report of Four Cases and Review. *Reviews of Infectious Diseases*, **13**, 1072-1076. https://doi.org/10.1093/clinids/13.6.1072

[14] Nguyen, V., Swartz, R.D., Reynolds, J., Wilson, D. and Port, F.K. (1987) Successful Treatment of Pseudomonas Peritonitis during Continuous Ambulatory Peritoneal Dialysis. *American Journal of Nephrology*, **7**, 38-43. https://doi.org/10.1159/000167427

[15] Forbes, B.A., Sahm, D.F. and Weissfeld, A.S. (2007) Bailey & Scott's Diagnostic Microbiology. 12th Edition, Mosby Elsevier, St. Louis.

[16] De Paoli Vitali, E., Rossi, M.R. and Farinelli, A. (1988) Pseudomonas-Like Species IIK-1 Peritonitis Intraperitoneal Dialysis. *Nephron*, **48**, 337. https://doi.org/10.1159/000184961

[17] Owen, J., Washco, V. and Reisin, E. (2016) Successful Return to Peritoneal Dialysis after a Case of Relapsing *Sphingomonas paucimobilis* Peritonitis. *Clinical Nephrology*, **86**, 287-289.

[18] Dervisoglu, E., Meric, M., Kalender, B. and Sengul, E. (2008) *Sphingomonas paucimobilis* Peritonitis: A Case Report and Literature Review. *Peritoneal Dialysis International*, **28**, 547-550. https://doi.org/10.1177/089686080802800523

[19] Lim, C.Y. and Tan, J. (2020) Case of Intra-Abdominal Abscess Due to *Sphingomonas paucimobilis* in a Patient on Peritoneal Dialysis: A Case Report and Review of Literature. *Indian Journal of Nephrology*, **30**, 196-200. https://doi.org/10.4103/ijn.IJN_271_19

[20] Li, P.K., Szeto, C.C., Piraino, B., de Arteaga, J., Fan, S., Figueiredo, A.E., Fish, D.N., Gofn, E., Kim, Y.L., Salzer, W., Struijk, D.G., Teitelbaum, I. and Johnson, D.W. (2016) ISPD Peritonitis Recommendations: 2016 Update on Prevention and Treatment. *Peritoneal Dialysis International*, **36**, 481-508. https://doi.org/10.3747/pdi.2016.00078

[21] Oki, R., Tsuji, S., Hamasaki, Y., Komaru, Y., Miyamoto, Y., Matsuura, R., Yamada, D., Doi, K., Kume, H. and Nangaku, M. (2021) Time Until Treatment Initiation Is Associated with Catheter Survival in Peritoneal Dialysis-Related Peritonitis. *Scientific Reports*, **22**, Article No. 6547. https://doi.org/10.1038/s41598-021-86071-y