Case Report

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*

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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

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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, aerobic 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,[2] 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

Table 1: Clinical Features of 14 Patients with *Sphingomonas paucimobilis* PD-Related Peritonitis (1984-2019)

Reference	Country	Age	PD vintage (month)	Previous peritonitis	Presenting complaints	Temperature at presentation (°C)	Etiology of ESRD
Glupczynski *et al*., 1984[7]	Belgium	74	6	NR	AP, CD, vomiting	36.6	Analgesic nephropathy
Glupczynski *et al*., 1984[7]	Belgium	33	13	Yes	AP, CD	Afebrile	Chronic pyelonephritis
Swann *et al*., 1985[8]	United Kingdom	61	33	Yes	CD	NR	Hypertensive nephrosclerosis
Baddour *et al*., 1985[9]	USA	50	36	No	CD 1st relapse: AP, CD 2nd relapse: AP, CD, vomiting	Afebrile	NR
Nguyen *et al*., 1987[10]	USA	65	28	Yes	NR	NR	Glomerulo-nephritis
De Paoli Vitali *et al*., 1988[11]	Italy	38	36	Yes	AP, CD, nausea	Afebrile	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	36.8	NR
Tambawala *et al*., 2011[14]	Pakistan	3.5	15	NR	CD, decreased oral intake, oral ulcers, perineal rash	Afebrile	Prune Belly syndrome
Lee *et al*., 2013[15]	Korea	63	72	Yes	AP, CD 1st relapse: 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 1st relapse: AP, CD	NR	Collapsing focal segmental glomerulo-sclerosis
Yılmaz *et al*., 2018[18]	Turkey	63	41	No	AP, CD, fever, vomiting	37.2	Hypertensive nephrosclerosis
Present case	Brunei	35	43	Yes	AP, CD	Afebrile	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	PD dialysate			Blood		
	WBC (per µL)	Initial Gram stain	Culture (day at which it turns positive)	WBC ($\times 10^9$/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[8]	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)			
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 bacillus	*P. paucimobilis* (D2)	NR	NR	NR
Dervisoglu *et al*., 2008[13]	148 (NE 90%)	No organism	*S. paucimobilis* (D3)	8.5	9.35	82
Tambawala *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*

Table 3: Treatment and Outcomes in 14 Patients with *Sphingomonas paucimobilis* PD-Related Peritonitis

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
		2) Amoxicillin PO (3g/day)	2) 7	Catheter removed on day 12
		3) After catheter removal, tobramycin IV (80 mg)	3) NR	Back on PD later
Swann *et al*., 1985[8]	Vancomycin IP (12.5 mg/L) + Gentamicin IP (4 mg/L)	Gentamicin IP (4 mg/L)	NR	Dialysate clear (day NR) Resolved
Baddour *et al*., 1985[9]	Cephalothin IP (1 g)	Cephalothin IP (250 mg/exchange)	4	Two episodes of relapses
	1st relapse: Cephalothin IP (1 g)	1st relapse: Cephalothin IP (250 mg/exchange)	1st relapse: 5	
	2nd relapse: Tobramycin IP (80 mg)	2nd relapse: Tobramycin IP (20 mg/exchange)	2nd relapse: 14	Resolved
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)
		2) Chloramphenicol (route NR)	2) 13	
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 Catheter removed (simultaneous new catheter insertion) 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)
		2) After catheter removal, imipenem IV	2) 7	Catheter removed on day 21
Tambawala *et al*., 2011[14]	Amikacin IP (2 mg/kg/bag)	1) Amikacin IP (25 mg/L)	1) 4	Resolved
		2) Meropenem IV (NR)	2) 7	
Lee *et al*., 2013[15]	Cefazolin IP (1 g/day) + Ceftazidime IP (1 g/day)	Cefazolin IP (1 g/day) + Ceftazidime IP (1 g/day)	14	Dialysate clear by day 3 Cloudy dialysate again on day 15
		1st relapse: Imipenem IP (1 g BD)	1st relapse:10	Catheter removed on day 17
Mohan *et al*., 2015[16]	Vancomycin IP (1 g single dose) + Ciprofloxacin IV (NR)	1) Tobramycin IP (4 mg/dL)	1) 21	Dialysate clear by day 5
		2) Meropenem IV (500 mg/day)	2) 21	Resolved
Owen *et al*., 2016[17]	Vancomycin IP (NR) + Ceftazidime IP (NR)	Ciprofloxacin PO (NR) + Ceftriaxone IP (NR)	21	Dialysate clear by day 4 Catheter removed (day NR)
		1st relapse: Ciprofloxacin PO (NR) + Ceftriaxone IP (NR)	1st relapse: 14	Back on PD after 6 weeks
Yılmaz *et al*., 2018[18]	Vancomycin IP (1 g single dose) + Ceftazidime IP (1 g/day)	Ceftazidime IP (1 g/day) + Amikacin IP (80 mg/day)	21	Dialysate clear by day 5 Resolved
Present case	Ampicillin/sulbactam IP (1.5 g BD) + Ceftazidime IP (1 g/day)	1) Ciprofloxacin IV (200 mg BD)	1) 35	Dialysate clear by day 10
		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.

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Conflicts of interest

There are no conflicts of interest.

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