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CASE REPORT

Sphingomonas paucimobilis Bacteremia in a Hemodialysis Patient and Literature Review

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Summary

Sphingomonas paucimobilis is an aerobic, non-fermentative gram-negative motile bacterium that may be an unusual infectious agent for immunocompromised host. Intravascular instrumentations are commonly applied in the hemodialysis patients. These procedures have an increased risk for infection with unusual, rare and opportunistic pathogens. *Sphingomonas paucimobilis* has been recently reported to cause community-acquired or healthcare-associated infections. A case of bacteremia associated with hemodialysis catheter caused by this bacterium that rarely leads to hospital infections has been represented.

Keywords

Bacteremia, Hemodialysis catheter, Chronic renal failure, *Sphingomonas paucimobilis*

Introduction

Sphingomonas paucimobilis is an aerobic, gram-negative, yellow-pigmented, non-fermentative, motile bacterium normally found in soil and freshwater which rarely causes serious life-threatening infections. These bacteria have been recently reported to cause community-acquired or healthcare-associated infections. This paper has aimed to draw attention to the fact that this agent may be found in hemodialysis patients even though it is rare by presenting a case of *Sphingomonas paucimobilis* bacteremia who have been receiving hemodialysis for 20 years.

Case

The 50-year-old female patient with end-stage renal disease, who had been receiving hemodialysis treatment for 3 days a week for last twenty years, has applied to emergency department of our hospital with complaints of chills-shivering and fever lasting for one month. Her anamnesis revealed chills-shivering that begin especially after hemodialysis procedure, fever of 37.5 °C by physical examination and findings of infection at entrance and tunnel of the right subclavian artery. System examinations showed no feature while laboratory tests results were as hemoglobin: 8.9 gr/dl, WBC: 3.600/mm^3, ESR: 91 mm/hour, CRP: 5.7 mg/dl (normal < 0.5 mg/dl). Her blood cultures obtained by two BacT/ALERT (Bio Merieux, France) bottles were incubated before empirical treatment of 2 gr cefazolin. Her blood culture grew methicillin-resistant coagulase negative staphylococci (CNS) and her treatment was switched to vancomycin 1 gr dose of every four days. Although, her complaints partially improved, blood culture tests were repeated due to the ongoing complaints of fever and chills for the last 15 days. In the third day of incubation, both blood cultures showed positive signal and a passage made through MacConkey and chocolate-like agar plates. Following 24-hour incubation at 35 °C, non-hemolytic, oxidase and catalase-positive gram-negative isolate grew in the chocolate agar plate.

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These gram-negative bacilli were identified *Sphingomonas paucimobilis* using VİTEK2 (Bio Merieux, France) automated identification system and were found resistant to ampicillin sulbactam, ticarcillin, piperacillin, piperacillin-tazobactam, meropenem, gentamicin, tobramycin and colistine; and sensitive to cefepime, tigecycline, trimethoprim-sulfamethoxazole and ciprofloxacin and moderately-sensitive to cefoperazone-sulbactam according to MIC results using the same system. First, the patient was initiated oral ciprofloxacin treatment as an ambulatory outpatient. However, then she was hospitalized and initiated IV ciprofloxacin 2 × 200 mg due to *Sphingomonas paucimobilis* growth in the repeated blood cultures at the 5th day of oral treatment. After her fever decreased and general condition improved at the 3rd day of hospitalization, the IV treatment was completed to 14 days and she was discharged.

Discussion

Infections are major mortality factors in the hemodialysis patients and these patients are always difficult problems that need to be solved in emergency departments. Many pathologies of lymphocyte and granulocyte function associated with uremia occur in these patients. Invasive procedures commonly performed during dialysis applications increase either predisposition to infections and incidence of rare infectious agents in these end-stage renal patients. *Sphingomonas paucimobilis* has been isolated first at 1977 as Pseudomonas paucimobilis and then classified currently as *Sphingomonas paucimobilis* (Group IIK, biotype 1) phylogenetically depending on its specific sphingolipid structure by Yabuuchi, et al. 1990 [1]. It is a non-fermentative, catalase and oxidase positive, Gram negative and very low motile bacillus with one single flagellum. Endotoxin-lipid A fraction of this bacterium has a sphingolipid-structure that stimulates mononuclear cells 105 times less than lipid A and causes secretion of TNF-α, interleukin-1 and interleukin-6. That explains the indistinct clinical symptoms [2]. *S. paucimobilis* infections are not had high mortality rates except of serious infections such as meningitis, septic shock syndrome or ventilator-associated pneumonia. These infections may be community-acquired or healthcare-associated. It has been shown that this agent could be found in also healthy people beside patients with immunosuppression, malignity and diabetes [3,4]. In Table 1, most of *S. paucimobilis* infections in the literature are summarized [3-14].

Table 1: Most of *S. paucimobilis* infections in the literature.

Infection	Source of infection	No. of patients	Country	Year
Septicaemia	N/A	1	USA	1979
Leg ulser	N/A	1	Australia	1979
Meningitis	N/A	1	UK	1979
Pseudo-infection	Contaminated irrigation fluid	4	USA	1981
Bacteremia	N/A	1	USA	1981
Nosocomial UTI	Contaminated water bottles	6	UK	1982
Bacteremia	N/A	1	Japan	1984
Peritonitis	Peritoneal dialysis	2	Belgium	1985
Peritonitis	Peritoneal dialysis	5	UK	1985
Peritonitis	Peritoneal dialysis	1	USA	1986
Bacteremia	N/A	1	USA	1987
Splenic abscess	N/A	1	USA	1988
Peritonitis	Catheter	1	Italy	1988
Brain abscess	Lawn dart	1	USA	1988
Empyema	N/A	1	USA	1990
Peritonitis	Hemodialysis fluid	1	UK	1990
Bacteremia	Hemodialysis fluid	1	USA	1991
Pseudomeningitis	N/A	1	USA	1991
Bacteremia, leg ulcer, cervical adenitis	Contaminated steril fluids	4	Spain	1991
Bacteremia	Bone marrow transplantation	1	USA	1991
Bacteremia-septic shock	N/A	1	USA	1992
Bacteremia	Catheter	2	USA	1992
Respiratory infection	N/A	2	Spain	1993
UTI	N/A	1	Spain	1994
Catheter related sepsis	Hemodialysis catheter	1	Australia	1994
Bacteremia	Catheter	2	Spain	1995
Bacteremia	Self-induced	1	Denmark	1995
Lung infections	Ventilators	85	Belgium	1996
Bacteremia	Catheter	4	Spain	1996
Bacteremia	Catheter	3	Germany	1998
Bacteremia	N/A	1	Spain	1998

Bacteremia	N/A	1	Spain	1998
Bacteremia, pneumoniae, wound infection, UTI, BTI	Indwelling devices	6	Taiwan	1998
Pneumoniae	N/A	1	Denmark	1998
Bacteremia	N/A	5	Spain	1998
Osteomyelitis	N/A	1	Spain	2000
Bacteremia	Catheter	1	Finland	2002
Bacteremia-septic arthritis	N/A	1	UK	2005
Endophthalmitis	N/A	1	UK	2006
Infection	Ventilator	47	Austria	2006
Pneumoniae	Ventilator	3	USA	2006
Bacteremia	N/A	4	Turkey	2007
Bacteremia	N/A	1	Saudi Arabia	2008
Peritonitis	Catheter	1	Turkey	2008
Bacteremia, diarrhoeal disease	Catheter	23	Korea	2008
Endophthalmitis	N/A	1	Korea	2008
Calf myositis	N/A	1	Italy	2008
Bacteremia	N/A	1	Turkey	2008
Bacteremia	Contaminated fentanyl	6	USA	2009
Pneumoniae	Ventilators	2	Turkey	2009
Bacteremia-septic arthritis	N/A	1	Taiwan	2009
Bromhidrosis	N/A	1	Italy	2009
Bacteremia	Distilled water	13	Turkey	2010
Bacteremia	N/A	16	Taiwan	2010
Peritonitis	Peritoneal dialysis	1	Pakhistan	2011
Bacteremia	N/A	1	Turkey	2011
Bacteremia, head-neck infection Pneumoniae, soft tissue infection meningitis, catheter-relted infections, UTI	N/A	55	Taiwan	2011
Septic arthritis	Intraarticular injection	1	Spain	2012
Meningitis	Wound	1	Malaysia	2013
Septisemi	Pyomyoma	1	Italy	2013
Osteomyelitis	N/A	1	Italy	2013
Bacteremia	N/A	1	Turkey	2013
Bacteremia	N/A	1	Turkey	2014

Table 2: The infection sources for *S. paucimobilis* according to reported articles up to now.

Infection type	**n**	**%**
Pneumoniae/lung infections	155	42.7
Bacteremiae/septicaemia	121	33.3
Soft tissue infections	16	4.4
Peritonitis	15	4.1
Head-neck infections	12	3.3
Catheter-related infections	11	3
Urinary tract infections	10	2.8
CNS infections	6	1.7
Osteomyelitis/septic arthritis	5	1.4
Gastrointestinal infections	4	1.1
Endophthalmitis	2	0.5
Others	6	1.7
Total	**363**	**100**

The agent has been isolated from a wide variety of clinical specimens including blood, urine, dialysate, pus, sputum, wound, sinovial and cerebrospinal fluid. Bacteremia/sepsis, ventilator-associated pneumonia, myositis, peritonitis, postoperative endophthalmitis, arthritis and surgical site infections have been reported. A meta-analysis evaluating *S. paucimobilis* infections between 1979 and 2010 years revealed that the most frequent clinical forms were bacteremia and peritonitis and there were 52 separate instances that the agent was isolated [3]. The infection sources for *S. paucimobilis* according to reported articles up to 2014 were summarized in Table 2. Many cases of *S. paucimobilis* bacteremia and peritonitis are resulted from contaminated solutions including distilled water, hemodialysis fluid and sterile drug solutions. Another review article from Taiwan evaluating totally 42 cases of *S. paucimobilis* bacteremia has shown that the malignity (57%), immunosuppression (41%), diabetes mellitus (12%) and end-stage renal disease (7%) were the most frequent co-morbid diseases [5]. In the same review, the researchers pointed out the high indwelling intravenous device rate of the cases (60%). Our case was also has been receiving hemodialysis for 20 years due to end-stage renal disease. In the literature, *S. paucimobilis* concomitance with Staphylococcus epidermidis has been reported in only one case [15]. Also in our patient, co-infection of CNS and *S. paucimobilis* may be considered. Actually, partially-improvement despite the proper therapy with vancomycin during the first bacteremia caused by CNS, could be related with dual bacteremia with CNS and *S. paucimobilis* before the hospitalization. Inability for

isolation of *S. paucimobilis* during the first bacteremia may be result from technical reasons or dominant and excessive growth of CNS bacteria hiding *S. paucimobilis* colonies in the culture plate.

S. paucimobilis is generally sensitive to tetracycline, co-trimoxazole, quinolones, and carbapenem. Its sensitivity to third generation cephalosporins and aminoglycosides is variable. In our case, the isolated agent was sensitive to only cefepime, cotrimoxazole, tigecycline and ciprofloxacin. Therapeutic failure to oral quinolone may be associated with lower bioavailability of drug or irregular oral therapy.

Consequently, intravascular instrumentations are commonly applied in the hemodialysis patients, development of immune response may be insufficient due to end-stage renal disease. These factors result in increased risk for infection with unusual, rare and opportunistic pathogens. *S. paucimobilis* should be kept in mind as a probable agent in dialysis patients with catheter-associated infection who experienced therapeutic failure.

Conflict of Interest

None to declare.

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