

Infective endocarditis related to the uncommon gram-negative pathogen *Sphingobacterium spiritivorum*

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Introduction

The generous *Sphingobacterium* consists of non-fermenting gram-negative environmental bacilli that are rarely involved in human infections. The species *Sphingobacterium multivorum* and *Sphingobacterium spiritivorum* have occasionally been associated with chronic respiratory infections [1], bacteremia [2-4], cellulitis [5] and peritonitis [4] in patients with severe underlying conditions. Here we present the first reported case of a patient who experienced an infective endocarditis related to *Sphingobacterium spiritivorum*.

A 61-year-old man with past medical history of nephrotic syndrome and hypertension was admitted in another hospital with acute kidney injury. Blood tests revealed serum creatinine 2.5 mg/dL and proteinuria 21.2 g/24 h. Ultrasonogram of kidneys and urinary tract was normal, and there was no evidence of calculi or hydronephrosis. A percutaneous kidney biopsy revealed focal segmental glomerulosclerosis. He was treated with prednisone (80 mg/day), spironolactone, losartan, atorvastatin, diuretics, and supportive treatments. Three weeks after first hospital discharge, the patient was readmitted in our nephrology service with an 8-day history of fatigue, mild mental confusion, drowsiness, asthenia, productive cough, and he denied fever and chills. Physical examination was remarkable only for crackles detected over the lower field of the right lung. Laboratory tests revealed serum creatinine 2.4 mg/dL, urea 182 mg/dL, sodium 139 mmol/L, and potassium 3.3 mmol/L; his hemoglobin was 10.0 g/dL, white blood cell count 4,600/uL (neutrophils, 86%; lymphocytes, 10%; monocytes, 4%), platelets $110 \times 10^9/L$ C-reactive protein 458 mg/dL, and serum albumin 1.4 g/dL. Urine dipstick was positive for blood (++) and protein (++++). Blood cultures were obtained, and he was started on empiric vancomycin and meropenem for concerns sepsis. The antibiotic was subsequently switched to intravenous oxacylin on day 5, as *Staphylococcus aureus* (MSSA) grew in the two sets of blood cultures obtained at admission. A transthoracic echocardiogram revealed no remarkable findings. The patient made remarkable improvement without any new or recurrent problems. After two weeks, computed tomography of the chest showed large right pleural effusions and numerous cavitary infiltrates in both lungs. Sample of pleural fluid and two blood cultures were positive for gram-positive cocci, which were later identified as methicillin-sensitive *Staphylococcus aureus* (MSSA). At this time, a new holosystolic murmurs (grade 4/6, heard best at the apex, and radiation to the carotids) was detected. The patient was hemodynamically stable, afebrile, and there were no skin or mucosal stigmata of endocarditis. Laboratory examination showed normocytic anemia (hemoglobin 10.3 g/dL), leukocytosis (total leukocyte count

10,800/uL) with neutrophilia (neutrophils 85%), and C reactive protein of 289 mg/L, and normal renal (serum creatinine 0.9 mg/dL) and liver function tests. A transoesophageal echocardiography revealed mitral valve prolapse, and an image suggesting mitral valve vegetation with moderate to severe grade regurgitation, and chordal rupture. Antibiotic (oxacylin) was changed to vancomycin and meropenem. During the follow up, patient's clinical picture deteriorated, and pulmonary rales progressed up to upper lung levels. The patient was then taken to the operating room emergently where he had a mitral valve replacement. Intraoperative histopathological results of the mitral valve indicated marked acute inflammation, fibroinflammatory debris, and presence of Gram-negative rod. After two days of incubation of mitral valve debris, non-motile, oxidase positive, non-fermenting Gram-negative bacilli grew, identified as *Sphingobacterium spiritivorum* (Vitek 2, bioMérieux, Marcy-L'Étoile, France), and confirmed using the Vitek MS -MALDI-TOF (Matrix Assisted Laser Desorption Ionization - Time of Flight, bioMérieux, Marcy-L'Étoile, France). Antimicrobial susceptibility testing was performed by automated microdilution broth method (Vitek 2). The results were reported as the minimal inhibitory concentration - MIC (CLSI Table 2B-5, M 100-S25-2015) [6], according to breakpoint criteria: ceftazidime (MIC < 4 µg/mL), cefepime (MIC < 4 µg/mL), imipenem (MIC < 0.5 µg/mL), meropenem (MIC ≤ 0.25 µg/mL), piperacillin/tazobactam (MIC < 8 µg/mL), ciprofloxacin (MIC ≤ 0.25 µg/mL), and sulfamethoxazole/trimethoprim (MIV MIC ≤ 20 µg/mL). Finally, like other flavobacteria-like organisms, it was highly resistant to colistin, amikacin and gentamicin. Piperacillin/tazobactam was associated to antibiotic treatment. He had no fever and his clinical picture was improved. Five weeks after surgical intervention, a new transoesophageal echocardiography did not show any evidence of vegetation or abscess. However, after two weeks the patient condition deteriorated, with fever, and haemodynamic instability. Blood cultures grew *Acinetobacter baumannii*. On the 40th day, the patient died of septic shock.

Discussion

We describe the first case of *Sphingobacterium spiritivorum* infective endocarditis in a native mitral valve with chordal rupture. Non-

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Received: August 06, 2019; Accepted: August 19, 2019; Published: August 21, 2019

Table 1. Cases of *Sphingobacterium spiritivorum* as human pathogen reported worldwide

	Marinella ⁷	Tronel ⁸	Koh ⁹	Kämpfer ¹⁰	PR
Year	2002	2003	2013	2005	2016
Sex	M	M	F	F	M
Age	72	84	68	34	61
Predisposing condition	Parkinson disease	Refractory anemia	Leukemia chemotherapy	NA	nephrotic syndrome
Symptoms	Fever Chills	Leg rash Fever	Fever	Dry cough Fever	Cough, fever Chills
Diagnosis	Cellulitis Sepsis	Cellulitis Sepsis	Sepsis	Extrinsic allergic alveolitis	endocarditis
Isolation	Blood	Blood	Blood	Steam iron Water reservoir	Mitral valve Infected debris
Treatment	NA	Amoxicillin-clavulanate 10 days	Ciprofloxacin 11 days	NA	Piperacillin-tazobactam 21 days
Outcome	Recovered	Recovered	Died 11 th day	Recovered	Died

M: male; F: female; NA: not available; PR: present report

fermenting gram-negative bacteria have been regarded historically as either environmental contaminants or organisms of low pathogenicity, and hence not clinically significant [2,4,7]. However, their emergence as significant causes of nosocomial bacteraemia has more recently been noted, particularly with regard to immunocompromised patients [2,3,7]. *Sphingobacterium* species, *S. multivorum* and *S. spiritivorum*, are non-fermenting, non-mobile, non-spore-forming aerobic gram-negative bacilli, and they are usually isolated from soil, water, and plant material.

S. spiritivorum has only been clearly incriminated as a human pathogen in four related cases worldwide. It was reported to cause of cellulitis and sepsis [8-10] or related to allergic reaction and hypersensitivity pneumonitis [11] (Table 1). In 2002, Marinella [8] first described the case of an elderly man with cellulitis and sepsis due to *Sphingobacterium spiritivorum* the patient most likely acquired from walking barefoot in his back yard. Tronel et al. [9] in 2003 reported a case of *S. spiritivorum* isolated from the blood cultures of an 84-year-old man with foot cellulitis. This bacterium was susceptible to most of the antibiotics tested, including glycopeptides, but was resistant to aminoglycosides and polymyxins; treatment with amoxicillin-clavulanate cured the infection. In 2013, Koh et al. [10] reported a case of *S. spiritivorum* bacteremia in a 68-yr-old woman, who was diagnosed with acute myeloid leukemia and subsequently received chemotherapy. The patient received antibiotic therapy for 11 days but died of septic shock. In 2005, Kämpfer et al. [11] reported a case of extrinsic allergic alveolitis caused by *S. spiritivorum* from the water reservoir of a steam iron. Although human infection is rare, *S. spiritivorum* can be a fatal opportunistic pathogen in immunocompromised patients [10]. More recently, Gupta et al. [12] reported a case of an 80-year-old woman with end stage renal failure, with type 2 diabetes mellitus, was established on long term haemodialysis; she experienced symptoms of a generalized infection with no specific signs on examination to identify a potential source. A set of blood cultures from the dialysis line and haemodialysis circuit both grew gram negative rods from the aerobic bottle only. The microorganism was identified to be *Sphingobacterium spiritivorum* [12].

Conclusion

We present a case of infective endocarditis caused by *S. spiritivorum*. The infection involved a native mitral valve and it was complicated by chordal rupture. The patient recovered following mitral valve replacement, and specific antibiotic therapy. However, in the following weeks the patient developed sepsis and died. Only a few case reports of human infections caused by *S. spiritivorum* have been published. To

our knowledge, this is the first reported case of infective endocarditis caused by *S. spiritivorum*. This bacterium was susceptible to most of the antibiotics tested, but was highly resistant to colistin, amikacin and gentamicin.

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