

Case Report

Sphingomonas paucimobilis infection in subcutaneous abdominal preservation of bone flap after craniotomy

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Abstract

Introduction: *Sphingomonas paucimobilis*, previously known as *Pseudomonas paucimobilis*, is a Gram-negative rod. It is emerging as an opportunistic pathogen that can infect individuals in community or hospital settings. It is believed that the natural habitat of this organism is soil and water, including water sources in the hospital environment.

Case report: We describe the case of a 46-year-old patient in whom *S. paucimobilis* was identified in the implanted bone flap after craniotomy. The postoperative bone flap was implanted in the right hypochondria and replaced after 8 weeks. There was a hypochondriac abscess in the area under the bone. Specimens from the thickened fascia and bone flap were cultured. The Gram stain showed Gram-negative rods and these rods were identified as *S. paucimobilis*. The patient was treated with a combination of Gentamicin 240 mg and Levofloxacin 750 mg once daily because the bacteria were resistant to carbapenem, trimethoprim-sulfamethoxazole, and anti-pseudomonal penicillin.

Conclusions: Although *S. paucimobilis* characteristically presents low virulence, for better patient management and outcome, the diagnosis should be immediately followed by appropriate antibiotic therapy guided by susceptibility test results of each case.

Key words: Craniotomy; flap; pathogen; preservation; *Sphingomonas*.

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Introduction

The genus *Sphingomonas* are aerobic Gram-negative, oxidase-positive, non-fermentative rods that are found in multiple environments [1]. They have been isolated from the natural environment, colonized water reservoirs, and soil [2,3]. *Sphingomonas paucimobilis* was first identified as a Centers for Disease Control and Prevention (CDC) Group IIk, biotype 1. In 1977, it was given its own taxonomic standing and given the name *Pseudomonas paucimobilis*. The bacteria were given their own genus, *Sphingomonas*, in 1990, and were selected as the genus type strain. The first *P. paucimobilis* infection was discovered in a sailor with a leg ulcer in 1979, and the bacterium was isolated from the wound material in pure culture. Many more cases of infection with the organism, as well as those colonized with the organism without presenting any symptoms, were described after that [4].

Sphingomonas may be misidentified by conventional identification systems [5]. The principal

distinctive biochemical characteristics of *P. paucimobilis* are its minimal growth requirements. It can be cultured on a variety of non-selective media, including blood and chocolate agar, but not on McConkey or media selective for enterobacteria. Colonies grown on blood agar are yellow-pigmented; however, this species is slow growing, and only small colonies may be observed after 24 hours of incubation. Growth occurs at 37 °C, but not at 42 °C, with optimal growth occurring at 30 °C. The name “*paucimobilis*” refers to difficulty experienced in demonstrating the motility of the organism, even in broth. Despite the presence of a polar flagella, only a few cells of a given population may be motile. Thus, motility is a difficult characteristic to demonstrate. Motility occurs at 18 °C to 22 °C, but not at 37 °C [4,6].

Initially, *S. paucimobilis* was thought to be a non-pathogenic environmental isolate [7]. *S. paucimobilis* can be widely found in the natural environment and has been isolated from water, soil, and other sources [2,3,8].

Humans can acquire *S. paucimobilis* in community or hospital settings [7]. Although infections associated with *S. paucimobilis* were reported to occur rarely, it has been more frequently reported in clinical settings [4].

Puca *et al.*, reported two cases of community-acquired primary bacteremia by *S. paucimobilis*; one of them was a septic shock in an immunocompromised patient suffering from diabetes mellitus for more than 10 years, and the other was a spondylodiscitis in an immunocompetent individual [8]. According to Lin *et al.*, most *S. paucimobilis* infections are healthcare-associated [3,9]. However, Toh *et al.* and Bayram *et al.*, reported that the incidence of community-acquired infections was higher than that of nosocomial cases [3,7,10]. It has been isolated from hospital environments such as distilled water, nebulizers, and multiple equipment used in medical care [4,8,11]. It has also frequently been isolated from hospital equipment (such as ventilators) and water sources [8,11].

It is seen in sporadic cases of various infections, such as catheter-related sepsis, primary bacteremia, pneumonia, meningitis, peritonitis, septic arthritis, and urinary tract infections. Small outbreaks can be caused by contaminated hospital devices and contaminated intravenous fluids. The presence of indwelling devices, an impaired immune system, and co-morbidities such as malignancy and diabetes mellitus are reported risk factors for *S. paucimobilis* infection [3].

S. paucimobilis is usually susceptible to carbapenems, aminoglycosides, trimethoprim-sulfamethoxazole, and piperacillin/tazobactam, but resistance patterns varied against cephalosporins and fluoroquinolones [3,7]. We report the case of a brain tumor patient with *S. paucimobilis* hypochondrium abscess that was resistant to the antibiotics. Most bone and soft-tissue infections were sensitive to carbapenems, aminoglycosides, trimethoprim-sulfamethoxazole, and anti-pseudomonal penicillin.

Case report

The 46-year-old female patient had a history of headaches in the past 2 years. During this period, there was a history of decreased consciousness. The patient suffered a daze and olfactory disorder 3 months earlier, but no impaired vision. On June 26, 2020, the patient was diagnosed with a frontal brain tumor. Craniotomy was performed on July 18, 2020, at the district general hospital. The postoperative bone flap was implanted in the right abdominal wall (hypochondria). There was no history of other diseases.

The patient was referred to our tertiary hospital on September 16, 2020, for brain tumor removal and was diagnosed with a frontal meningioma. Initial physical examination was within normal limit, Glasgow Coma Scale (GCS) score was E4M6V5, body mass index 28.4 kg/m², heart rate of 90 beats per minute, blood pressure of 135/84 mmHg, respiratory rate of 18 breaths per minute, and no episode of fever (36.9 °C). Hematology parameters were within normal range as follows: hemoglobin 12.7 g/dL (range 12-14), leukocyte count 9.8 x 10⁶/mm³ (range 5-10×10⁶, differential counts showed 1% eosinophils, 1% stab neutrophils, 60% segmented neutrophils, 36% lymphocytes, and 2% monocytes), and thrombocyte count 383×10⁶/mm³ (range 150-400×10⁶). The patient was screened for HBsAg and HIV, and the results were negative. Due to the pandemic, the patient was also tested for the SARS-CoV-2 using Reverse Transcription Polymerase Chain Reaction (RT-PCR), and the result was negative. Chest radiology and electrocardiography were within normal range.

The patient was given cefazolin 2 g as a prophylactic antibiotic before tumor removal surgery on September 18, 2020. The abdomen was opened after tumor removal to remove the implanted bone flap. There was yellow fluid in the area under the bone suggesting a hypochondrium abscess. After debridement, the external ventricular drain was attached to the abdomen. Specimens from the thickened fascia and bone flap were cultured. The post-surgery laboratory parameters showed: normochromic normocytic anemia (hemoglobin 11.2 g/dL, range 12-14), leukocytosis (leukocyte count 23.7×10⁶/mm³, range 5-10×10⁶) with neutrophilia (differential counts showed 1% eosinophils, 1% stab neutrophils, 60% segmented neutrophils, 36% lymphocytes, and 2% monocytes), and thrombocyte within normal range (thrombocyte count 332×10⁶/mm³, range 150-400×10⁶). The patient was given ceftriaxone 2 g twice daily, and metronidazole 500 mg every 8 hours.

The thickened fascia and bone flap specimens were incubated on thioglycolate broth and yielded positive growth after 24 hours of aerobic incubation at 37 °C. The Gram stain showed Gram-negative rods. Isolates from thioglycolate broth were incubated on blood agar and yielded positive growth after 24 hours of aerobic incubation at 37 °C. The colonies were yellowish, had no detectable hemolysis, and showed Gram-negative rods. All the isolates were examined by Vitex® 2 Compact (bioMérieux, Lyon, France), and these rods were identified as *S. paucimobilis*. The antibiotic susceptibility of *S. paucimobilis* found in thickened

fascia was similar to that in the bone flap. Antibiotics susceptibility was performed by Vitex® 2 Compact (bioMérieux, Lyon, France). It was susceptible to gentamicin, amikacin, ciprofloxacin, and tigecycline. The results of thickened fascia and bone flap cultures are detailed in Table 1.

On September 23, 2020, histopathological examination showed a meningotheliomatous and transitional type of meningioma, categorized as World Health Organization (WHO) grade 1. The hematology parameters were hemoglobin 12.3 g/dL (range 12-14), leukocyte $18.4 \times 10^6/\text{mm}^3$ (range $5-10 \times 10^6$), and thrombocyte count $383 \times 10^6/\text{mm}^3$ (range $150-400 \times 10^6$). Differential counts showed 84% segmented neutrophils, 8% lymphocytes, and 8% monocytes. As laboratory results came out on the 5th-day post-surgery, antibiotic therapies were changed according to the results. The patient was treated with gentamicin 240 mg and levofloxacin 750 mg once daily, and metronidazole 500 mg every 8 hours, for 7 days. On the 7th-day post-surgery, the drain was removed.

Discussion

Repair of the cranial bone defect is important for several reasons: 1) for brain protection, to start an early rehabilitative program; 2) to reconstruct the original cranial cerebral compartment and some authors suggest that an early cranioplasty may have a better outcome; 3) for aesthetic and psychological issues related to the patients. Different materials have been proposed to achieve a better reconstruction, from autologous bone to various heterologous materials. Autologous bone is the simplest, cheapest, and most suitable material that can be used, but it has to be adequately preserved. Traditionally, after craniotomy, the bone flap was

frozen, sterilized, and replaced after weeks or months [12].

In our case, the bone flap was implanted in the right hypochondria, and might have become devitalized later, leading to hypochondrium abscess as complications. The *S. paucimobilis* that caused the abscess in our case might be endogenous or unknown. Charity and Foukas [13], reported the case of a 16-year-old male who had both osteomyelitis and septic arthritis, whereas Puca et al. [8], reported the case of spondylodiscitis due to *S. paucimobilis* infection. *S. paucimobilis* is an opportunistic pathogen and identification of the organism from the clinical specimen is rare [8]. *S. paucimobilis* nosocomial infections can be caused by the patient's previous colonization, the environment (through the implantation of various indwelling devices), or the contamination of sterile fluid in the hospital. In the majority of cases, the cause was unknown [14].

According to Demiray et al. [3], the most common concomitant diseases were malignancy (33.33%) and diabetes mellitus (30.30%). The presence of an indwelling catheter (72.73%) and hospitalization in the intensive care unit (66.67%) were the most common risk factors. Diabetes mellitus and alcoholism were not determined as risk factors for *S. paucimobilis* infection in our case.

Although Gram-negative infections frequently result in significant morbidity and mortality, *S. paucimobilis* infections seem to show a low rate of complications and a favorable prognosis. It is possible that this low virulence is caused by the presence of atypical lipopolysaccharide in the outer membrane, a deficiency of bacterial endotoxins, and a different enzyme profile [15]. The bacterium has been isolated from many patients with no signs or symptoms attributable to infection due to this organism. Despite the majority of the patients exhibiting a good prognosis with no reported complications resulting from appropriate antimicrobials, some required a more aggressive therapy. The most complicated cases occurred in immunocompromised hosts, but other healthy individuals were also affected. The diagnosis should be immediately followed by the appropriate antibiotic therapy. However, the susceptibility and resistance patterns of *S. paucimobilis* varied widely across the reported cases and studies, which highlight the need for individualized case-by-case management [4].

In general, they are significantly resistant to cephalosporin, especially the third generation, and to penicillin [15]. Demiray et al. [3], reported tigecycline

Table 1. Culture and resistance test result.

Specimen	Thickened fascia	Bone flap
ISOLATE	<i>Sphingomonas paucimobilis</i>	<i>Sphingomonas paucimobilis</i>
Susceptibility		
Gentamicin	S*	S
Trimethoprim/Sulfamethoxazole	R#	R
Piperacillin/Tazobactam	R	R
Cefazolin	R	R
Ceftriaxone	R	R
Ceftazidime	R	R
Amikacin	S	S
Ciprofloxacin	S	S
Cefepime	R	R
Meropenem	R	R
Tigecycline	S	S

* S: sensitive; # R: resistant.

was most frequently susceptible, but there are no data in the literature for comparison. *S. paucimobilis* was also found to be sensitive to imipenem, meropenem, netilmicin, and amikacin. Bayram et al. and Toh et al. [10,11] reported that fluoroquinolones, carbapenems, and trimethoprim/sulfamethoxazole were the most effective antibiotics. Lin et al. [9], stated that *S. paucimobilis* was frequently sensitive to fluoroquinolones, carbapenems, and beta-lactam/beta-lactamase combinations. Besides, cephalosporins and beta-lactam/beta-lactamase were found to be less effective. The antimicrobials most commonly used for *S. paucimobilis* infections were carbapenems and third-generation cephalosporin/aminoglycoside combinations [3].

Most bone and soft-tissue infections are sensitive to carbapenems, aminoglycosides, trimethoprim-sulfamethoxazole, and anti-pseudomonal penicillin. In our case, it was resistant to carbapenem, trimethoprim-sulfamethoxazole, and anti-pseudomonal penicillin, but susceptible to aminoglycoside, fluoroquinolone, and tigecycline. The patient was treated with Gentamicin 240 mg and Levofloxacin 750 mg once daily, and Metronidazole 500 mg every 8 hours. Some researchers suggest combinations of third-generation cephalosporin/aminoglycoside for the treatment of this type of infection. However, standardized therapies cannot be established at present, because of the different antimicrobial susceptibility patterns in previous studies. Therefore, relying on antimicrobial susceptibility testing results is the most logical and appropriate approach [3].

Conclusions

S. paucimobilis is an emerging pathogen. The nosocomial or community-acquired *S. paucimobilis* infection is related to previous comorbidities. Although *S. paucimobilis* characteristically presents low virulence compared with other Gram-negative bacteria, the diagnosis should be immediately followed by appropriate antibiotic therapy. Since antibiotic resistance varies, treatment should be guided by susceptibility test results of each case for better patient management and outcome.

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