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Sphingomonas paucimobilis meningitis in an immunocompetent patient

Meningitis por Sphingomonas paucimobilis *en un paciente inmunocompetente*

The genus *Sphingomonas* belonging to the *Sphingomonadaceae* family (phylum Alphaproteobacteria), contains 139 species (https://lpsn.dsmz.de/genus/sphingomonas). However, only *Sphingomonas paucimobilis* (*S. paucimobilis*) and *Sphingomonas parapaucimobilis* (*S. parapaucimobilis*) are considered clinically relevant. *S. paucimobilis* has been found in water and soil and is considered an opportunistic pathogen.[1] However, it can cause infections in immunocompetent individuals. It has been detected mainly in intravenous catheters, mechanical ventilators, nebulizers, contaminated solutions, and dialysis devices in hospital settings.[1] We present a case of meningitis due to this unusual microorganism; only six cases in adult patients have been reported to date.[2–7] Our patient had hydrocephalus as a complication; nevertheless, he had a satisfactory recovery.

A 30-year-old, previously healthy male student with no risk factor for infectious diseases, presented with headache and vertigo that persisted for 7 days. He received an otorhinolaryngological evaluation and was treated for suspected bacterial pharyngitis with penicillin without improvement. A few days later, the patient started with behavioral disturbances, and neck pain. His vital signs upon admission were as follows: blood pressure: 140/80 mmHg, pulse: 102 bpm, and temperature: 37.2 °C; a neurological examination revealed neck stiffness.

A blood test showed the following results: white cell count, 14.2×10^9/L; neutrophils, 13.5×10^9/L; lymphocytes, 1.3×10^9/L; hemoglobin, 15.4 g/L; platelets, 252×10^9/L; erythrocyte sedimentation rate, 35 mm/h; C-reactive protein, 6.92 mg/dL. Blood chemistry test results showed no significant alterations; serological tests for human immunodeficiency virus, hepatitis B and C viruses, and urine toxicology tests were negative.

Empiric antibiotic therapy was prescribed, with ceftriaxone 4 g daily, vancomycin 30 mg/kg/day, and acyclovir 10 mg/kg as meningoencephalitis was suspected. Cranial computed tomography (CT) results did not show relevant findings, and to rule out other sources of infection, a thoracoabdominal CT was performed, which was normal. After a lumbar puncture, the following were observed: clouded fluid; white blood cell count, 165/mm^3; neutrophils, 97%; glucose, 11 mg/dL; protein, 1110.2 mg/dL. Cerebrospinal fluid (CSF) India ink staining was negative; however, Gram-staining showed gramnegative rods. Therefore, the antibiotic treatment was changed to meropenem (6 g daily). The CSF was seeded on sheep blood agar and after 48 h, yellow-pigmented flat colonies with irregular, watery, and slippery shapes with a tendency to converge were observed. Microbial identification using VITEK®2 Compact 60 with the GN ID card reported *S. paucimobilis*. The susceptibility test was performed by the VITEK®2 Compact 60 with the GN69 susceptibility card, based on the breakpoints of the current Clinical and Laboratory Standards Institute document M100,[8] showing the following results: resistance to ceftriaxone, cefepime, and aztreonam with minimal inhibitory concentration (MIC) ≥64 μg/mL; susceptibility to piperacillin/tazobactam (≤4/4 μg/mL), meropenem (≤0.25 μg/mL), amikacin (≤2 μg/mL), gentamicin (≤1 μg/mL), tobramycin (≤1 μg/mL), ciprofloxacin (1 μg/mL), tigecycline (≤0.5 μg/mL), and trimethoprim/sulfamethoxazole (≤1/20 μg/mL).

On hospitalization day 8, the patient's condition deteriorated, and he developed symptoms suggestive of intracranial hypertension. CT imaging of the brain revealed hydrocephalus, requiring a subsequent ventriculoperitoneal shunt. After 2 weeks of antibiotic treatment, another lumbar puncture was performed. CSF cultures were negative. He was discharged from the hospital after a 14-day-long course of meropenem. His mental function returned to normal, and he was sent for physical rehabilitation. After 5 weeks of follow-up, he was asymptomatic and reinstated to his normal activities.

S. paucimobilis is a strictly aerobic, gramnegative bacillus with weak oxidase and catalase activities, and has slow motility because of a single polar flagellum. *S. paucimobilis* grows well on non-selective media, such as blood agar and chocolate agar, although it does not grow on MacConkey agar.[9] After 24 h of incubation on a blood agar medium, their colonies are small, approximately 1 mm in diameter, convex, smooth, and produce a yellow insoluble pigment. Notably, its susceptibility to vancomycin is unusual in gramnegative non-fermenting rod-shaped bacteria. This microorganism has been isolated from blood, sputum, urine, bile, and CSF, among others, in patients with risk factors, such as malignancy, chronic kidney disease, diabetes mellitus, immunosuppressive drug usage, and alcoholism.[10]

Our patient did not have any risk factors associated with *S. paucimobilis* infection; nevertheless, as *S. paucimobilis* is ubiquitous in soil and water, they could be likely sources. The patient's clinical presentation suggested meningitis without other foci of infection, while most other reported cases of *S. paucimobilis* menin-

Table 1
Demographic and clinical characteristics adult patients with meningitis caused by *S. paucimobilis* in previously reported cases.

Age and sex	Comorbidity	Likely source	Clinical manifestations[a]	Csf	Image	Culture and sensitivity	Treatment	Outcome
30-M (Our case)			Headache, stiff neck, vertigo, no fever	WBC: 165/mm^3 neutrophils 97%, Glucose: 11 mg/dL, Proteins: 1110.2 mg/dL	CT: Hydrocephalus	CSF: *S. paucimobilis* susceptible: meropenem	Initial: Ceftriaxone, vancomycin and acyclovir. **Meropenem 14 days.** Ventriculoperitoneal shunt	Recovery
66-F Bae SW et al. (2021)[5]	Breast cancer treated with chemotherapy (Immunocompetent)	Acupuncture	Fever, drowsiness, neck rigidity	WBC: 429/μL. red blood cells: 100/μL, glucose: 11 mg/dL, protein 369.51 mg/dL	Initial MRI without implications. CT third day of hospital stay: Ventricular Enlargement (Hydrocephalus)	CSF: *S. paucimobilis* susceptible: cefotaxime, ciprofloxacin, ceftazidime and cefepime. Blood culture: *L. monocytogenes*, susceptible: ampicillin and vancomycin	Initial: Vancomycin ceftazidime and dexamethasone. **Ampicillin/sulbactam and ceftriaxone for 21 days.** External ventricular drainage. Ventriculoperitoneal shunt	Recovery
50-F Mehmood et al. (2018)[3]	Rheumatoid arthritis		Headache, dizziness, chills, neck pain, stiffness, no fever	WBC 5/mm^3, neutrophils 4%. red blood cells: 95/mm^3, glucose: 60 mg/dL, protein 37 mg/dL	MRI and CT without alterations	*S. paucimobilis* susceptible to meropenem	**Meropenem 21 days**	Recovery
48-F Göker et al. (2017)[2]			Sudden onset speech disturbance, worsening of the general condition. GCS 5/15, no fever, no evidence of meningeal irritation	Bloody CSF and high protein content	4 × 4 cm hemorrhagic lesion in the basal ganglia. (Hydrocephalus)	*S. paucimobilis*: susceptible to meropenem and gentamicin.	External Ventricular Drainage. Ceftriaxone prophylaxis. **Meropenem**	Death
39-F Bolen et al. (2015)[6]	End-stage renal disease and deceased donor kidney transplant. In treatment with tacrolimus, mycophenolic acid, and prednisone		Severe headache, dizziness, nausea, neck pain, impaired gait. Short-term memory deficit, Bilateral Babinski sign. Meningism. Fever	WBC: 781/mm^3 (64% neutrophils). Red blood cells: 9/mm^3. Proteins: 2789 mg/dL. Glucose 18 mg/dL	MRI: diffuse hyperintensity in T2 along the lateral ventricles and the third ventricle	*S. paucimobilis* susceptible to meropenem	Initial: Vancomycin, ceftriaxone, ampicillin, and ganciclovir. **Meropenem 3 weeks**. Mechanic ventilation	Recovery
31-M Tai ML et al. (2014)[4]		Farmer. Leg wound	Fever, headache, fatigue, weight loss. Lethargy, speech disturbances. GCS: 10/15. Neck stiffness, Hyporeflexic pupils	WBC: 210/mm, Glucose 3.1 mmol/L, Protein 2.47 g/L	CT: Brain edema and hydrocephalus	*S. paucimobilis* susceptible to ceftriaxone, ceftazidime, ciprofloxacin, and piperacillin/tazobactam	**Ceftriaxone and acyclovir. Anti-tuberculosis.** Mechanic ventilation	Death
39-M Hajiroussou et al. (1979)[7]	Epilepsy		Seizures, headache, fever, stiffness, kerning sing (+)	WBC: 200/mm^3		*S. paucimobillis*	Streptomycin, rifampicin, isoniazid for 5 days	Recovery

CSF, cerebrospinal fluid; CT, computed tomography; WBC, white blood cell count; CSF, cerebrospinal fluid; CT, computed tomography; MRI, magnetic resonance; Highlighted in bold, definitive treatment after CSF culture.

[a] Three cases, including ours, did not present with fever.

gitis did not show another source and presented the classic triad of meningitis (i.e., fever, nuchal rigidity, and altered mental status). Among the six previously reported cases of *S. paucimobilis* meningitis, two presented without fever, and three out of five cases with comorbidities had underlying conditions predisposing them to infections (Table 1).[2,3] Among the complications of bacterial meningitis, the following stand out: ischemic and hemorrhagic events (14–37%), altered mental state (80%), hydrocephalus (3–21%), and brain abscess (5%), with altered mental state and hydrocephalus presented in our patient. In 50% of the reported cases of *S. paucimobilis* meningitis, the most common complication was hydrocephalus,[2,4,5] and only one patient had ventriculitis.[6]

S. paucimobilis produces beta-lactamase; therefore, it is usually resistant to first-generation penicillins, such as piperacillin, and cephalosporins, such as cephalothin.[11] In approximately 31% of the cases, resistance to cefepime and ceftazidime has been reported.[1] In some series, the resistance rate to ciprofloxacin was 58%[11]; *S. paucimobilis* is usually susceptible to carbapenems, trimethoprim/sulfamethoxazole (TMP/SXZ), ampicillin/sulbactam, and piperacillin/tazobactam.[1,9,10] The susceptibilities reported in our case are consistent with those of previous reports, and monotherapy with carbapenems, aminoglycosides, and TMP/SXZ are good options for empiric therapy. Most patients were treated for 21 days; however, our patient showed improvement and negative results for bacterial culture test 14 days after starting treatment with meropenem. The mortality rate of *S. paucimobillis* has been reported to be only 5.5%.[9] The absence of lipopolysaccharide in the outer capsule and the lack of endotoxin activity may explain the low virulence,[12] resulting in a favorable prognosis in most cases.[9–12]

Although *Sphingomonas* spp. infections are rare, there has been an increase in reported cases in recent years and must therefore be considered even in immunocompetent patients. The treatment is based on appropriate empirical antimicrobial choice, appropriate adjustments based on susceptibility tests, and supportive therapy in the event of complications, which yields good outcomes in most cases.

Informed consent and patient details

Informed consent was obtained from the patient.

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Conflict of interest

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