First Report of Sphingomonas koreensis as a human pathogen in a patient with meningitis

Marbjerg, Lis H; Gaini, Shahin; Justesen, Ulrik S

Published in: Journal of Clinical Microbiology

DOI: 10.1128/JCM.03069-14

Publication date: 2015

Document version: Final published version

Citation for published version (APA):

Terms of use
This work is brought to you by the University of Southern Denmark through the SDU Research Portal. Unless otherwise specified it has been shared according to the terms for self-archiving. If no other license is stated, these terms apply:

• You may download this work for personal use only.
• You may not further distribute the material or use it for any profit-making activity or commercial gain
• You may freely distribute the URL identifying this open access version

If you believe that this document breaches copyright please contact us providing details and we will investigate your claim. Please direct all enquiries to puresupport@bib.sdu.dk
First Report of *Sphingomonas koreensis* as a Human Pathogen in a Patient with Meningitis

Lis H. Marbjerg,a,b Shahin Gaini,a Ulrik S. Justesen
c
Department of Infectious Diseases, Odense University Hospital, Odense, Denmark; Department of Clinical Microbiology, Veje Hospital, Veje, Denmark; Department of Clinical Microbiology, Odense University Hospital, Odense, Denmark

*Sphingomonas koreensis* is an aerobic Gram-negative rod originally described in 2001 following isolation from natural mineral water in Korea. Here, we report a case study with *Sphingomonas koreensis* as the causative agent of meningitis. To our knowledge, this is the first documented case of *Sphingomonas koreensis* as a human pathogen.

CASE REPORT

In 1997, a 14-year-old female patient underwent surgery due to a syrinx in the cervical portion of her medulla. A cisternoperitoneal shunt system was inserted, and the column was stabilized with pedicled screws and metal plates corresponding to T1-T2 and T11-T12.

In 2003, she was diagnosed with secondary amenorrhea due to a pituitary adenoma. A transsphenoidal resection of the tumor was attempted in 2005, but bleeding resulted in the operation being unsuccessful.

In November 2012, the patient was diagnosed with bacterial meningitis. The cerebrospinal fluid (CSF) showed the following: elevated leukocytes of 290 nucleated cells/μl with 80% neutrophils, protein concentration at 0.7 g/liter, and CSF glucose concentration of 1.1 mmol/liter. No bacteria were observed in a concentrated Gram stain of the CSF, but *Staphylococcus epidermidis* and *Staphylococcus warneri* were isolated. After susceptibility testing of the two isolates, the patient received a 2-week course of intravenous ceftriaxone (4 g once daily). Twelve days after stopping the ceftriaxone treatment, the symptoms of meningitis returned. The CSF showed elevated leukocytes of 853 nucleated cells/μl with 85% neutrophils, protein concentration at 0.6 g/liter, and CSF glucose concentration of 0.6 mmol/liter. Again, no bacteria were observed in a concentrated Gram stain, but *S. epidermidis* was cultured from the CSF and intravenous ceftriaxone was reinstituted. Initially, the clinical response was satisfactory; however, the symptoms recurred in January 2013. Following this recurrence, the decision was made to change the treatment plan to intravenous meropenem (2 g three times a day) and vancomycin (1 g twice daily). Treatment with intravenous meropenem and vancomycin continued until the removal of the shunt system in February 2013. It should be noted that the peritoneal part of the shunt system could not be removed. The remaining part had no interaction with the central nervous system.

Two weeks after surgical removal of the shunt, the patient was admitted to the Department of Infectious Diseases with a headache, stiff neck, and confusion. The CSF showed the following: elevated leukocytes of 335 nucleated cells/μl with 70% neutrophils, protein concentration of 1.2 g/liter, and CSF glucose concentration of 0.6 mmol/liter. Treatment with intravenous meropenem (2 g three times a day) and vancomycin (1 g twice daily) was initiated, and temporary external lumbar drainage was performed. A magnetic resonance imaging scan showed a cranialosal fistula at the point of the previous transsphenoidal resection from 2005. Subsequently, a brain computed tomography showed two osseous defects in the sphenoid sinus. Blood cultures taken at the time of admission were without growth, but *Sphingomonas koreensis* was cultured from the CSF. The CSF was taken before initiation of antibiotic treatment, and no bacteria were observed in a concentrated Gram stain. Additional CSF cultures taken from the lumbar drain over a 3-day period in late February and March did not result in growth. However, 5 days following hospitalization, *S. koreensis* was cultured again from the CSF. Treatment with intrathecal gentamicin (8 mg once daily) was added to the intravenous meropenem and vancomycin treatment, and the lumbar drain was replaced. Cultivation of the tip of the drain was without growth. After antimicrobial susceptibility testing, the antibiotic treatment was changed to oral trimethoprim-sulfamethoxazole (80/400 mg twice daily). The two osseous defects in the sphenoid sinus were surgically repaired during the hospitalization period. Slowly, the lumbar drainage was reduced, which concluded with removal of the drain and discharge of the patient in late March 2013. On discharge, she had received a total of 3 weeks of treatment with oral trimethoprim-sulfamethoxazole.

In September 2013, the patient was readmitted to the Department of Infectious Diseases presenting with fatigue, headache, and neck pain that had been increasing in severity over 3 to 4 days. The patient described the symptoms as being similar to those of the previous episode of meningitis. The clinical examination revealed no neck stiffness, and she was fully conscious. Her temperature was 37.5°C. The CSF showed 45 nucleated cells/μl, of which 27 were neutrophils. The protein concentration was normal at 0.34 g/liter, but the glucose concentration was low at 1.6 mmol/liter compared to a serum glucose of 6.3 mmol/liter. Laboratory results
rent symptoms of meningitis. To date, which is more than a year, the patient has had no recur-

trimethoprim-sulfamethoxazole treatment, she received another

ing side effects (fatigue and nausea). After a total of 3 weeks of oral

was suspected that the dosage used for the previous episode had

ingitis with higher doses of oral trimethoprim-sulfamethoxazole

were growth (yellow colonies) was observed in a concentrated Gram stain of the CSF taken at

The two isolates of S. koreensis from February and September

is motile with a single polar flagellum and catalase, oxidase, and

data were obtained from the two spinal fluid samples on 5% horse blood agar after 72

ment of meningitis.

The second isolate was also subjected to partial 16S rRNA gene

obtained an unambiguous diagnosis of Sphingomonas koreensis

The genus Sphingomonas was first proposed in 1990. It is char-

The genus Sphingomonas has since been divided into 4 genera: Sphingomonas sensu stricto, Sphingobium, Novosphingobium, and Sphingopyxis (2). S. koreensis was originally described in 2001, after it was dis-

was first proposed in 1990. It is char-

S. koreensis is considered to be a low-virulence organism, and mortal-

FIG 1 Sphingomonas koreensis on 5% horse blood agar.
In conclusion, the unique symptoms of meningitis clearly indicates that this was a true pathogen and not a contaminant. In this case, the finding of S. koreensis as the causative organism, and this could explain why the patient was asymptomatic in February 2013 or through the two osseous defects in the sphenoid sinus. It could have been introduced during the removal surgery of the cystoperitoneal shunt system in February 2013 or through the two osseous defects in the sphenoid sinus. S. koreensis must be considered to be a low-virulence organism, and this could explain why the patient was asymptomatic for several months between February and September 2013. Furthermore, when she presented at our department in September 2013, it was with relatively mild symptoms, and she had no fever. The diagnosis of meningitis was made because the patient was known to have recurrent meningitis and because the patient described the symptoms as being similar to those of the previous episode. The exact origin of the S. koreensis is unknown. As a commensal of the environment, including natural mineral water, there could be multiple sources.

This case demonstrates the importance of species identification, even if isolates are considered or suspected to be contaminants. In this case, the finding of S. koreensis at the second episode of meningitis clearly indicates that this was a true pathogen and the cause of meningitis in the patient. In conclusion, the unique case we report here indicates that antibiotic therapy should be directed against the specific organism, S. koreensis.

ACKNOWLEDGMENTS

We thank Amanda G. Vang for proofreading the manuscript and Olav D. Larsen for assistance with the work.

REFERENCES