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Otomastoiditis caused by *Sphingomonas paucimobilis*: case report and literature review

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ABSTRACT

Sphingomonas paucimobilis is an aerobic Gram-negative bacillus that, although rare in humans, most commonly infects immunocompromised and hospitalized patients. Among the 59 pediatric cases of *S. paucimobilis* infection reported in the literature, the most common diagnosis involves isolated bacteremia. These cases are related to sporadic or epidemic infections. Death related to this infection occurred in only one case. The authors report a case of an 11-year-old boy with the diagnosis of Sphingomonas paucimobilis otomastoiditis and a thorough review of the literature on this infection in pediatrics. The patient presented a 20-day history of fever, otalgia, otorrhea, and progressive retroauricular swelling with protrusion of the left ear; despite 15 days of amoxicillin regimen. His past medical history included chronic bilateral otitis media, but no cause of immunosuppression was found. A brain computed tomography scan showed left otomastoiditis associated with a large circumscribed fluid collection with deep involvement of the soft tissues of the temporal region, including the subperiosteal space. Blood tests showed neutrophilia and elevated C-reactive protein. Surgical manipulation of the cited collection drained a large amount of a fetid purulent secretion. Ceftazidime and clindamycin were empirically initiated. The outcome was favorable, with fever defervescence and resolution of the scalp deformation. Culture of the drained secretion was positive for *S. paucimobilis*. Ciprofloxacin was scheduled for a further 10 days after discharge. The follow-up showed complete recovery. As far as we know, this is the first case of *S. paucimobilis* otomastoiditis, complicated with subperiosteal abscess in an immunocompetent child. The authors call attention to the increasing number of reports on *S. paucimobilis* infection over the years, and therefore to the importance of this pathogen, which was previously underestimated.

Keywords

Humans; Child; Sphingomonas; Gram-negative Bacterial Infections; Immunocompromised Host.

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INTRODUCTION

Infection by *Sphingomonas paucimobilis* is rare. Although it occurs mostly in immunocompromised patients, cases of infected immunocompetents have been reported. We present a case of subperiosteal abscess due to a complicated otomastoiditis in an immunocompetent child. No previous case of subperiosteal abscess or otomastoiditis caused by *S. paucimobilis* has been reported. *S. paucimobilis* infection reports have been increasing in pediatric cases, encouraging us to conduct a literature review accompanied by the analysis of the clinical, demographic, and treatment data of the retrieved pediatric case report. We searched the PubMed and Google Scholar databases using the terms "*Sphingomonas paucimobilis*", "*Pseudomonas paucimobilis*," "Sphingomonas," and "paucimobilis"; checking out the references of each elected article for any paper that could have been missed. All available publications related to *S. paucimobilis* infection in children, from 0 to 18 years, were retrieved.

To the best of our knowledge, this is the first review of pediatric infections due to these bacteria.

CASE REPORT

An 11-year-old boy was brought to the emergency department with a 20-day history of fever, otalgia, otorrhea, and progressive retroauricular swelling, displacing the left ear anteriorly and downwards (Figure 1).

He was previously prescribed amoxicillin for 15 days without improvement. He lived in an orphanage and his HIV serology was negative. He had a previous diagnosis of bilateral chronic otitis media, but no other comorbidity was present. A computed tomography (CT) scan was performed (Figure 2) showing left otomastoiditis associated with a large fluid collection involving the subperiosteal layer of the temporal bone and the surrounding soft tissues, measuring 10.1 × 6.0 × 2.2 cm. Blood tests showed a white blood count of 22.4 × 10^3mm^3 (reference value [RV]: 4.5-11.0 × 10^3/mm^3)(neutrophils 81.2%; RV: 40-70%) and C-reactive protein of 142 mg/L (RV: < 5 mg/L).

Surgical intervention on the retroauricular mass found a large amount of fetid and purulent secretion, a sample of which was sent off for microbiology studies. Ceftazidime and clindamycin were empirically started. The outcome was successful immediately after the surgical procedure, with fever defervescence, and normalization of blood tests. After 8 days of incubation, the culture of purulent secretion was positive for *S. paucimobilis*, but unfortunately, the antibiogram could not be tested because the bacteria in the culture media was no longer available. Therefore, based on literature data, we decided to maintain the initial antibiotic regimen and discharged the patient on the tenth day of hospitalization, scheduling 10 more days of oral ciprofloxacin to be taken at home.

METHODS

We searched PubMed, Google Scholar and LILACS databases using the terms "Sphingomonas paucimobilis", "Pseudomonas paucimobilis", "Sphingomonas" and "paucimobilis". The references of each selected article were checked out in order to

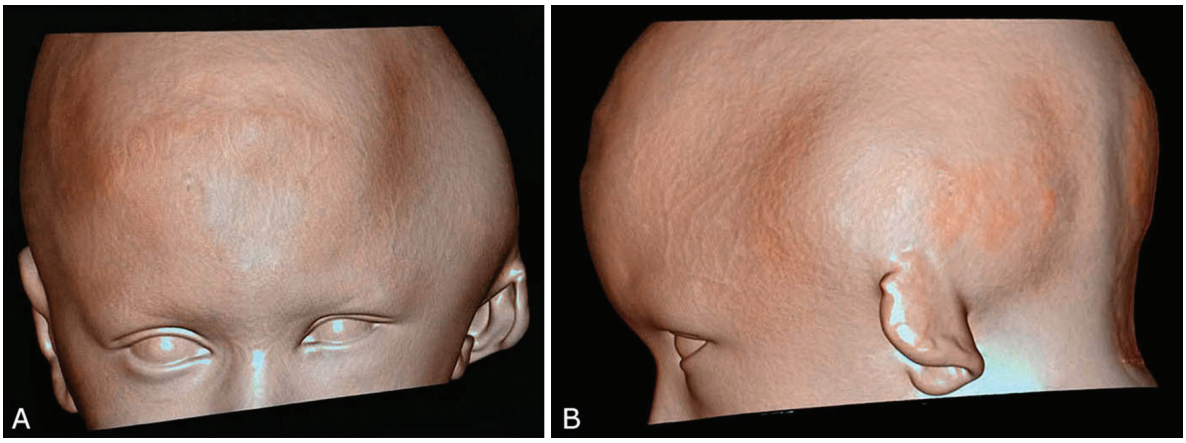

Figure 1. 3D reformation of the mastoid computed tomography showing the bulging tumoral mass displacing the left ear downwards and anteriorly.

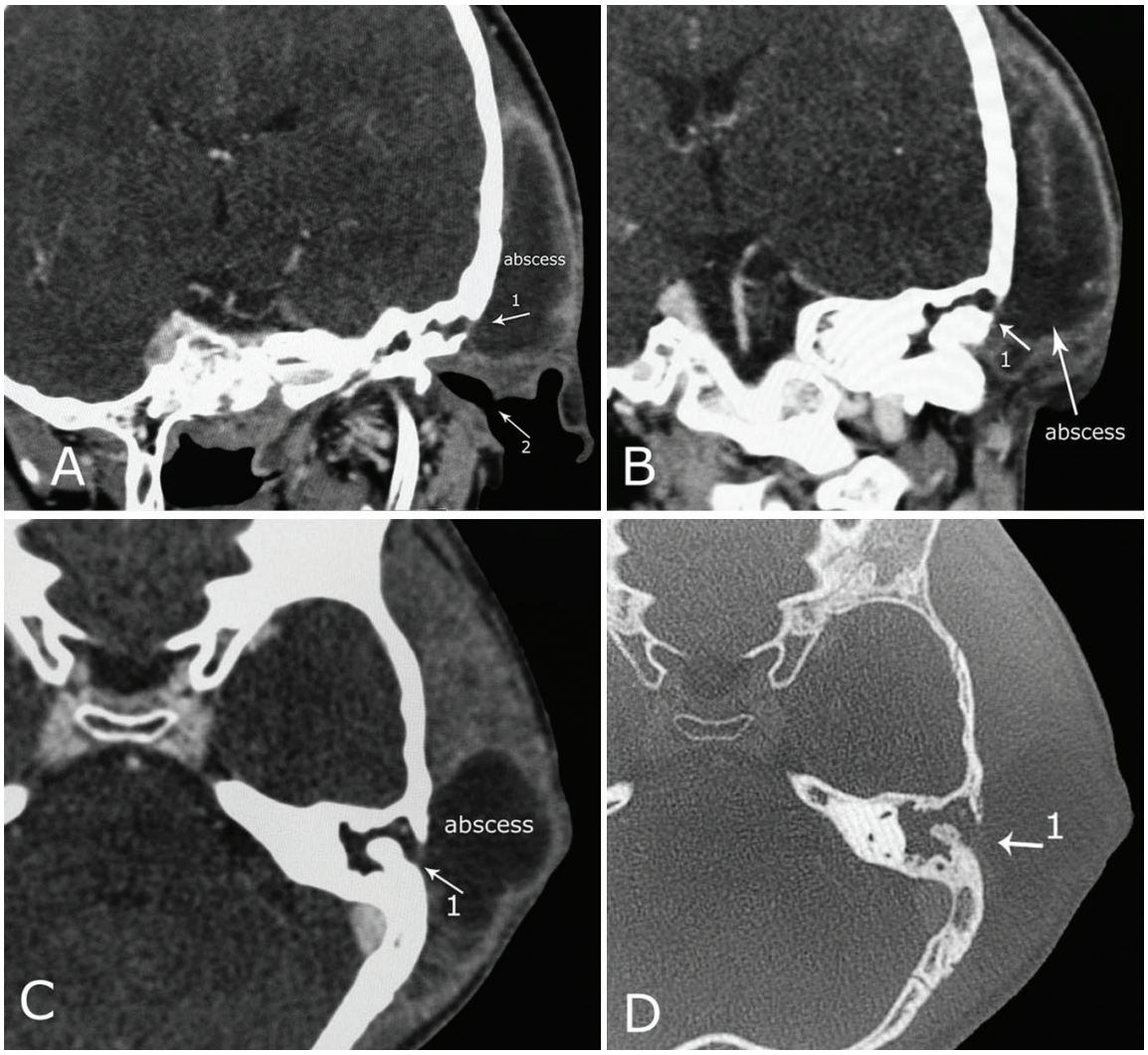

Figure 2. Computed tomography of the mastoid**. A** and **B** - Coronal plane. **C** and **D** - Axial plane. **D** represents the bone window. Arrow 1 shows the erosion of the temporal bone; arrow 2 the external ear canal. The abscess is connected with the mastoid do to the erosion of the temporal bone. The collection involved all soft tissues of the temporal region limited externally by the skin.

avoid missing any other paper. From a total of 1699 articles, 20 were related to *Sphingomonas paucimobilis* infection in children and adolescents up to 18 years, and therefore were selected.

DISCUSSION

The genus *Sphingomonas* comprises 13 Gram-negative, non-fermentative, non-sporing, oxidase- and catalase-positive bacilli. *Sphingomonas paucimobilis* is slightly motile (thus paucimobilis) and forms yellow pigmented colonies in blood agar.[1] It was believed that *S. paucimobilis* was the only specimen of clinical importance; however, recently *Sphingomonas mucosissima* and *Sphingomonas adhesiva* also have been related to human infection.[2,3] These bacilli are ubiquitous in nature (especially in water and soil) and in nosocomial environments causing mild-to-severe community and nosocomial illnesses.[1,4]

First described by Holmes et al.[5] in the 1970s as *Pseudomonas paucimobilis*, it was later reclassified as *Sphingomonas paucimobilis* in accordance with phylogenetic data by Yabuuchi et al.[6] in the 1990s. Curiously, this bacteria was related to the death of the coral reef on the coast of Florida, USA.[7]

The full-automated microbiology identification systems like "autoSCAN-W/A" and the "Vitek GNI1," may fail to identify some genera and species. Sung et al.[8] showed that the major misidentified bacteria were the Gram-negative non-fermentative, especially *S. paucimobilis* in the autoSCAN-W/A system.

In 1979, the case of an infected leg ulcer of a Japanese seaman in Australia, was the first reported human infection.[9] In 1981, the first pediatric colonization case by this bacteria was reported,[10] but only in 1988 did Tiffany and Kline[11] describe the first pediatric case of *S. paucimobilis* infection related to a brain abscess in a 6-year-old boy after a head trauma. Since then, the reports on this infection has increased, totaling 59 reported cases in pediatrics to date. Before 1980, no *S. paucimobilis* infection in children was reported. One case was reported between 1981 and 1990, three cases between 1991 and 2000, 28 cases between 2001 and 2010, and 27 cases between 2011 and March 2014, including ours (Figure 3). Despite the reported cases of infections, numerous cases of colonization by this bacteria were also published.[10,12,13]

Table 1 lists all elected cases of pediatric *S. paucimobilis* infection retrieved in our review. Table 2 summarizes the demographic and clinical data. The median age was 4.4 years (ranging from 3 days to 17 years); 22 cases occurred during the neonatal period (37.3%). The observed male:female ratio was 1.7:1.0. Hospital-acquired disease was identified in 66.1% and previous comorbidities were present in all of those patients.

The most common infection site was primary bacteremia (74.6%) followed by catheter-related bloodstream infection (10.2%), urinary tract infection (3.4%) and one case (1.7%) of each of the following: peritonitis related to continuous ambulatory peritoneal dialysis, cervical adenitis, central nervous system (CNS) infection, CNS abscess, osteomyelitis/septic arthritis, pneumonia, and otomastoiditis. Prematurity was the commonest associated comorbidity observed in 16 cases (41%); 13 cases were associated with malignancy (33%), out of which all were from the hematopoietic system; 2 cases (5.2%) occurred in children with Down syndrome, 2 cases (5.2%) with gut malformations, and 6 cases associated with other isolated diseases.

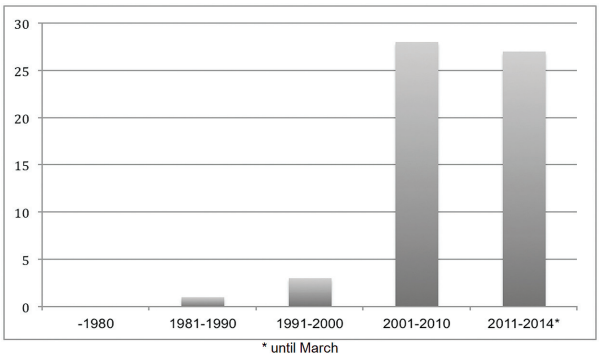

Figure 3. The number of reported pediatric cases of *S. paucimobilis* infection from 1980 until March 2014.

S. paucimobilis has been isolated from blood, sputum, urine, wound, ascitic liquid, cerebrospinal fluid, synovial liquid, and purulent secretion (brain, bone, and mastoid).[11,14-23]

Some studies associate *S. paucimobilis* outbreaks with contamination of the nosocomial environment; however, none of the reviewed papers demonstrated the same strain of the contamination with those of the infection site.[17,18,20,24] Consequently, the endogenous source of the infection is the current theory on the route of infection.[17,23] In addition, infection was also related to hospitalization, indwelling devices, and previous comorbidities, but our own case and other recent studies show that community-acquired infections are more prevalent than previously thought.[19,25]

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.[16,19,21,24,25] Only one case of pediatric death has been reported related to *S. paucimobilis* infection, which occurred in a premature neonate with septic shock.[20] It is supposed 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.[6,26,27]

S. paucimobilis antibiotic susceptibility varies according to the study. In general, they are significantly resistant to cephalosporin, especially the third generation, as well as to penicillins. Lin et al.[19] showed that all isolated bacteria produced β-lactamase; however, other studies showed a resistance to cephalosporins of around 20%.[21,22] There is also reported resistance to aminoglycosides, quinolones, trimethoprim, and sulfamethoxazole, and lesser to carbapenems.[4,14-18,21,22] In previous studies, the recommendation of initial antibiotic therapy was a third generation cephalosporin added to an aminoglycoside; however, more current studies suggest that carbapenems or quinolones show a better susceptibility pattern.[4,16,17,22]

Despite supposed initial inappropriate antibiotic therapy reported in some cases of the literature, none presented any complication due to the delayed institution of the appropriate treatment.[21,22] Since antibiotic resistance varies, treatment should be guided by the susceptibility test results of each case.[4,16]

Table 1. Cases of *S. paucimobilis* infection in children, since the first publication in 1988 until March 2014

Condition	Age	Gender	Underlying conditions	Therapy	Country	Source of outbreak	Number of patients	Reference
CR-BSI	14y	-	Acute myeloid leukemia	Trimethoprim-Sulfamethoxazole	Saudi Arabia	Hospital	1	14
Osteomyelitis/ septic arthritis	16y	M	ALL	Amikacin/Ceftazidime	United Kingdom	**	1	15
Bacteremia	<1y	F	Chylothorax	Not described	Korea	Hospital	1	16
Bacteremia	2y	F	Aplastic anemia	Not described	Korea	Hospital	1	16
Bacteremia	8y	M	ALL	Not described	Korea	Hospital	1	16
Bacteremia	<1y	M	Neonatal sepsis	Not described	Korea	Hospital	1	16
CAPD peritonitis	14y	F	End-stage renal disease	Not described	Korea	Hospital	1	16
CR-BSI	1y	M	Anaplastic ependymoma	Not described	Korea	Hospital	1	16
CR-BSI	17y	M	Ewing sarcoma	Not described	Korea	Hospital	1	16
CR-BSI	10d	M	Prematurity/RDS	Not described	Taiwan	Hospital	1	17
Bacteremia	6y	F	Lymphoma	Imipenem/Teicoplanin	Turkey	Hospital	1	18
Bacteremia	5y	F	Neuroblastoma	Imipenem/Teicoplanin	Turkey	Hospital	1	18
Bacteremia	11y	M	Non-Hodgkin lymphoma	Imipenem/Teicoplanin	Turkey	Hospital	1	18
Bacteremia	3y	M	Acute myeloid leukemia	Imipenem/Teicoplanin	Turkey	Hospital	1	18
Pneumonia	5y	M	None	Ampicillin-Sulbactam	Taiwan	Community	1	19
UTI	5m	M	None	Ampicillin/Gentamicin	Taiwan	Community	1	19
Bacteremia*	0m	-	Prematurity	Not described	Turkey	Hospital	13	20
Bacteremia	2y	M	None	Ceftriaxone	India	Community	1	20
Bacteremia	7y	F	None	Cefuroxime	Turkey	Community	1	22
Bacteremia	1m	F	None	Ampicillin/Sulbactam	Turkey	Community	1	22
Bacteremia	15y	F	None	Cefotaxime	Turkey	Community	1	22
Bacteremia	1y	F	None	Cefuroxime	Turkey	Community	1	22
Bacteremia	8y	F	Neutropenia-ALL	Piperacillin/Tazobactam	Turkey	Hospital	1	22
Bacteremia	8d	F	Prematurity	Meropenem/Vancomycin	Turkey	Hospital	1	22
Bacteremia	12y	F	None	Piperacillin/Tazobactam	Turkey	Hospital	1	22
Bacteremia	27d	F	None	Meropenem	Turkey	Hospital	1	22

ALL = acute lymphoblastic leukemia); CAPD = continuous ambulatory peritoneal dialysis; CNS = central nervous system; CR-BSI = catheter-related bloodstream infection; PSAGN = steroid-induced immunosuppression due to glomerulonephritis; RDS = respiratory distress syndrome; UTI = urinary tract infection; *= case of fatal outcome; **= the author could not define if it was a community or hospital infection.

Table 1. Continued...

Condition	Age	Gender	Underlying conditions	Therapy	Country	Source of outbreak	Number of patients	Reference
Bacteremia	3y	M	None	Cefotaxime	Turkey	Community	1	22
Bacteremia	12y	M	PSAGN	Cefuroxime	Turkey	Community	1	22
Bacteremia	18d	M	Prematurity	Ampicillin/Amikacin	Turkey	Community	1	22
Bacteremia	9y	M	Down syndrome	Meropenem/Vancomycin	Turkey	Community	1	22
Bacteremia	1m	M	None	Ampicillin/Sulbactam	Turkey	Community	1	22
Bacteremia	3d	M	None	Ampicillin/Amikacin	Turkey	Community	1	22
Bacteremia	23d	M	None	Ampicillin/Sulbactam	Turkey	Community	1	22
Bacteremia	1y	M	Imperforate anus	Ceftazidime	Turkey	Hospital	1	22
Bacteremia	23d	M	Duodenal atresia	Cefotaxime/Amikacin	Turkey	Hospital	1	22
Bacteremia	11y	M	None	Meropenem	Turkey	Hospital	1	22
Bacteremia	4y	M	Neutropenia-ALL	Piperacillin/Tazobactam	Turkey	Hospital	1	22
Bacteremia	5y	M	Neutropenia-ALL	Piperacillin/Tazobactam	Turkey	Hospital	1	22
CNS infection	10y	M	None	Cefotaxime/Vancomycin	Turkey	Community	1	22
CR-BSI	4y	M	Burn injury	Meropenem/Amikacin	Turkey	Hospital	1	22
CR-BSI	10y	F	Neutropenia-ALL	Piperacillin/Tazobactam	Turkey	Hospital	1	22
UTI	4m	F	None	Cefuroxime	Turkey	Community	1	22
Bacteremia	1y	M	Cardiopathy-Down syndrome	Piperacillin/Tazobactam	Turkey	Hospital	1	4
Otomastoiditis	11y	M	None	Ceftazidime/Clindamycin/ Ciprofloxacin	Brazil	Community	1	Present Study
Bacteremia	6y	M	None	Cefotaxime/Gentamicin	Spain	Community	1	23
Cervical adenitis	8y	M	None	Ampicillin	Spain	Community	1	23
Brain abscess	6a	F	None	Ampicillin/Chloramphenicol	USA	Community	1	11

ALL = acute lymphoblastic leukemia); CAPD = continuous ambulatory peritoneal dialysis; CNS = central nervous system; CR-BSI = catheter-related bloodstream infection; PSAGN = steroid-induced immunosuppression due to glomerulonephritis; RDS = respiratory distress syndrome; UTI = urinary tract infection; *= case of fatal outcome; **= the author could not define if it was a community or hospital infection.

Table 2. Demographic and clinical characteristics of *S. paucimobilis* infection in children—data of the literature review

Condition	No. of cases	Mean age (range)	Gender (ratio) M:F	Healthy/previous comorbidity (no. of cases)	Nosocomial/Community-acquired (No. of cases)
Isolated bacteremia*	44	3.1y (0-15y)	1.5:1	13/31	31/13
CR-BSI	6	8.3y (0-17y)	4:1	0/6	6/0
UTI	2	4.5m (4-5m)	1:1	2/0	0/2
CAPD peritonitis	1	14y	0:1	0/1	1/0
Cervical adenitis	1	8y	1:0	1/0	0/1
CNS infection	1	10y	1:0	1/0	1/0
CNS abscess	1	6y	0:1	1/0	0/1
Osteomyelitis/septic arthritis	1	16y	1:0	0/1	***
Pneumonia	1	5y	1:0	1/0	0/1
Otomastoiditis**	1	11y	1:0	1/0	0/1
Total	**59**	**4.4y (0-17y)**	**1.7:1**	**20/39**	**39/20**

CAPD = continuous ambulatory peritoneal dialysis; CNS = central nervous system; CR-BS = catheter-related bloodstream infection; UTI = urinary tract infection; *= case of fatal outcome; **= present study; ***= the author could not define if it was nosocomial or community acquired infection.

In conclusion, *S. paucimobilis* is an emerging pathogen in pediatric patients, and not just a contaminant of the hospital environment. The nosocomial or community-acquired infection is related to previous comorbidities, which are mostly associated with immunosuppression due to prematurity and malignancy. However, previously healthy patients may also be involved. Compared with other Gram-negative bacteria, *S paucimobilis,* characteristically present low virulence; however, death has been reported already in one pediatric case. Notwithstanding the resistance to beta-lactam antibiotics, the first choice of treatment consists in carbapenems or quinolones.

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