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Short Communication

Meningitis Due to S*phingomonas paucimobilis* in a Pediatric Patient: A Case Report

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Abstract

Sphingomonas paucimobilis is a gram-negative, nonfermentative, aerobic, oxidase and catalase positive, non-spore producing bacterium characterizied with yellow pigment production and motile with polar flagella. In this study, we describe an unusual case of *S. paucimobilis* meningitis in a patient with ventriculoperitoneal shunt. A 13-years-old girl was brought to the emergency room with complaints of fever and headache for two days. She had ventriculoperitoneal shunt surgery following posterior fossa tumor surgery 10 years before. Upon physical examination, the patient was uncomfortable and had 39°C body temperature. Laboratory results were as follows: hemoglobin, 12.6 g/dL; leukocyte count, 18,800/mm^3 (76% neutrophil); platelet count, 179,000/mm^3; and C-reactive protein, 34 mg/dL. The patient was prediagnosed with meningitis and cerebrospinal fluid sample (CSF) was taken before the empirical vancomycin and meropenem treatment. Shunt was removed, CSF was drained externally. The sample was sent to the Microbiology laboratory. CSF sample was inoculated on 5% sheep blood agar and Eosin-Methylene Blue agar. After 24 hours of incubation at 37^{o}C, Gram-negative bacilli were grown on media. The isolate was identified as *S. paucimobilis* using the VITEK 2 automated system. The bacterium was susceptible to imipenem, colistin, levofloxacin, meropenem and cefepime. The patient was treated successfully with appropriate antibiotic treatment. We describe this unusual case of ventriculoperitoneal shunt infection with *S. paucimobilis.* In conclusion, *S. paucimobilis* is an infectious agent that is prevalent in nature but may also be isolated in the hospital setting. It can lead to nosocomial or community acquired infections. Although it can be eliminated with prophylactic therapy, sensitivity pattern should be definitely studied to determine the optimal treatment.

Keywords: Shunt infection, *Sphingomonas paucimobilis,* Cerebrospinal fluid, Meningitis

Резюме

Sphingomonas paucimobilis е Грам-отрицателна, неферментативна, аеробна, оксидаза- и каталаза-положителна, не-спорообразуваща бактерия, характеризираща се с продуцирането на жълт пигмент, подвижна, с полярни флагели. В настоящото изследване се съобщава за необичаен случай на менингит, причинен от *S. paucimobilis*, в пациент с вентрикулоперитонеален шунт. Момиче на 13-години постъпва в спешното отделение с оплаквания за треска и главоболие от два дни. Тя е с вентрикулоперитонеален шунт след операция на мозъчен тумор преди 10 години. След първоначалния преглед пациентката е в дискомфорт, с телесна температура 39°C. Лабораторните резултати са: хемоглобин 12.6 g/dL; левкоцити 18,800/mm^3 (76% неутрофили); тромбоцити 179,000/mm^3 и С-реактивен протеин 34 mg/dL. Предварителната диагноза е менингит и е взета проба от цереброспинална течност (ЦСТ) преди емпиричното третиране с ванкомицин и меропенем. Шунтът е отстранен, а ЦСТ се дренира външно. Изпратена е проба в микробиологичната лаборатория. ЦСТ е инокулирана върху 5% овчи кръвен агар и еозин-метилен блу агар. След 24 ч. инкубация при 37 ^{o}C, върху средите се развиват Грам-отрицателни бацили. Чрез автоматизирана система VITEK 2

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изолатът е идентифициран като *S. paucimobilis.* Бактерията е чувствителна към имипенем, колистин, левофлоксацин, меропенем и цефепим. Пациентката е третирана успешно с подходящи антибиотици. В статията описваме този необичаен случай на инфекция на вентрикулоперитонеален шунт с *S. paucimobilis*. В заключение, *S. paucimobilis* е инфекциозен агент, широко разпространен в природата, но освен това може да бъде изолиран и в болнична среда. Може да причини инфекции, свързани с медицинското обслужване или придобити в общността. Въпреки че може да бъде елиминиран с профилактична терапия, за оптималното третиране е важно да се проучва лекарствената му чувствителност.

Introduction

Sphingomonas paucimobilis is a non-fermentative, obligately aerobic, gram negative, non-spore producing bacillus and motile by a single polar flagellum. It is oxidase, catalase and esculine hydrolysis positive, urease and indole negative and characterized with a yellow pigment on sheep blood agar. The bacterial growth requires minimum 24-48 hours of incubation, at 30-37°C (not at 42°C) and 5% CO_2 or ambient atmosphere. Bacterial growth occurs nearly 48 hours on sheep blood agar (Smalley *et al.,* 1983; Yabuuchi *et al*., 1990; Ryan and Adley, 2010; Tai and Velayuthan, 2014; Tille, 2014).

S. paucimobilis is rarely isolated from human materials and has a limited role as a causative agent. Because it is rarely encountered in clinical settings there is limited information about its epidemiology and ability to cause human infection. According to recent literature, although *S. paucimobilis* has minor clinical importance immune-compromised patients, diabetes mellitus and alcoholism are considerable risk factors for primary bacteremia. Infection, especially in immune compromised patients, can lead to septic shock (Tille, 2014) . *S. paucimobilis* has been isolated from many clinical specimens such as wound infections (brain abscess, splenic abscess, leg ulcer), urine, vaginal, cervical samples, blood cultures and cerebrospinal fluid (CSF). In the literature, there are a few reported cases of meningitis caused by *S. paucimobilis* (Ryan and Adley, 2010; Tai and Velayuthan, 2014; Tille, 2014) . In this study, we describe an unusual case of *S. paucimobilis* meningitis in a patient with ventriculoperitoneal shunt. This case is reported to emphasize that *S. paucimobilis* should be kept in mind as a nosocomial infectious agent and the infections should be treated according to the susceptibility test results.

Case Presentation

A 13-year-old girl was brought to the emergency room with complaints of fever and headache for two days. She had had ventriculoperitoneal (VP) shunt following posterior fossa tumor surgery 10 years before. Upon physical examination, the patient was uncomfortable and had 39°C body temperature. Laboratory results were as follows: hemoglobin, 12.6 g/dL; leukocyte count, 18 800/mm^3 (%76 neutrophil); platelet count, 179 000/mm^3; and C-reactive protein, 34 mg/dL. The patient was prediagnosed with meningitis and CSF sample was taken before the empirical vancomycin and meropenem treatment. Shunt was removed, CSF was drained externally. On different days, triplicated CSF samples were sent to the Microbiology laboratory and inoculated on to 5% sheep blood agar and Eosin-Methylene Blue agar (bioMérieux, France). After 24-48 hours of incubation at 37°C, yellow pigmented, slow growing, catalase and oxidase positive, urease and indol negative Gram-negative bacilli grew on media. The same bacteria were isolated in all three samples. The isolates were identified as *S. paucimobilis* using the VITEK 2 (Biomerieux, France) automated system. Antimicrobial susceptibility test was carried out with VITEK 2 automated system and the results were evaluated according to the European Committee on Antimicrobial Susceptibility Testing (EUCAST) break point for *Pseudomonas sp* (EUCAST, 2016). The bacteria were susceptible to imipenem (MIC 2 µg/mL), colistin (MIC< 0.5 µg/mL), levofloxacin (MIC 0.5 µg/mL), meropenem (MIC 0.5 µg/mL) and cefepime (MIC 8 µg/mL). Although the patient was being treated in the clinic, in the third week CSF culture reproduction continued with *S. paucimobilis*. Levofloxacin was added to the treatment, two weeks after CSF sterilization. Shunt revision was performed again. The patient was treated successfully with appropriate antibiotic treatment. Therefore, we present this unusual case of VP shunt infection with *S. paucimobilis.*

Discussion

S. paucimobilis was first described in 1977 as *Pseudomonas paucimobilis* and was changed in the 1990s to its recent name (Yabuuchi *et al*., 1990). Bacteria can be found in soil and water but can proliferate in distilled water, hemodialysis flu-

ids and sterile drug solutions. So it can cause both community-acquired and nosocomial infections. *S. paucimobilis* has minor clinical significance and its virulance is low so the bacterium is usually not checked in routine hospital analysis, but it may be clinically important for immuno-compromised patients, diabetes mellitus, post operative and nosocomial infection (Tille, 2014). Therefore, it should be considered as a causative agent alternative to the *Pseudomonas*, *Stenotrophomonas* and *Burkholderia* species for these patients (Ryan and Adley, 2010; Tille, 2014).

Recently, there have been many studies about this bacterium, which show that the importance of *S. paucimobilis* has increased over the years (Maragakis *et al*., 2009; Ryan and Adley, 2010; Deveci *et al*., 2017). Bacteraemia, ventilator-associated pneumonia, myositis, peritonitis, postoperative postoperative endophthalmitis and catheter-related infections have been reported in literature (Ryan and Adley, 2010; Tille, 2014). But meningitis caused by *S. paucimobilis* have been reported only in a few case (Hajirousou *et al*., 1979; Tai and Velayuthan, 2014; Bolen *et al*., 2015; Deveci *et al*., 2017). Ryan *et al*. (2010) have evaluated 240 case reports published about *S. paucimobilis* and have discovered 52 different cases of infection relating to the presence of *S. paucimobilis*. In the study, they reported 20 cases of bacteraemia/sepsis, five cases of peritonitis, three cases of pneumonia, three cases of urinary tract infection and the others in only one case. In our country, Erdem *et al*. (2010) have reported a case of surgical-site surgical-site infection owing to *S. paucimobilis*. Basoglu *et al*. (2013) isolated 11 *S. paucimobilis* strains from tracheal secretion samples (7), urine (2), wound (1) and blood culture (1). Bulut *et al*. (2008) reported a case of hospital acquired bloodstream infection caused by *S. paucimobilis.*

In the literature, Hajiroussou *et al.* (1979) first reported a case of meningitis in a 39-year-old patient, caused by *P. paucimobilis* (recently *S. paucimobilis*), and since that time there have been a few cases of meningitis caused by *S. paucimobilis* (Tai and Velayuthan, 2014; Bolen *et al*., 2015; Deveci *et al*., 2017). Deveci *et al*. (2017) have reported a case of community-acquired *S. paucimobilis* meningitis in an adolescent patient. The study was the first case of adolescent meningitis caused by *S. paucimobilis*. Tai and Velayuthan (2014) have reported *S. paucimobilis* meningitis in a 31-year-old farmer who was working in soil and had a wound in the leg. They reported that the bacteria entered his body through the wound resulting in bacteraemia and later meningitis. In another study, Bolen *et al*. (2015) presented the case of *S. paucimobilis* meningitis in an immuno-compromised patient, which was also the first reported case of ventriculitis caused by this bacterium. In this study, we describe an unusual case of *S. paucimobilis* meningitis in a 13-years old patient with ventriculoperitoneal shunt.

As yet, there is no standard procedure for antibiotic susceptibility of *S. paucimobilis*. Based on *in vitro* susceptibility studies, case specific therapy indication was required, because antibiotic resistance is reported differently in many articles (Fink, 2009). In some studies antibiotic resistance of *S. paucimobilis* is reported according to EUCAST breakpoint breakpoint for *Pseudomonas* (Deveci *et al.*, 2017). In our study, we evaluated antibiotic resistance according to EUCAST *Pseudomonas* break-point breakpoint (EUCAST, 2016). This case is reported to emphasize that *S.paucimobilis* should be kept in mind as a nosocomial infectious agent and the infections should be treated according to the sensitivity test results.

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