

Case Report

A Rare Cause of Bacteremia in a Pediatric Patient with Down Syndrome: *Sphingomonas Paucimobilis*

Mehmet Özdemir¹✉, Sevgi Pekcan², Mehmet Emin Demircili¹, Fatma Esenkaya Taşbent¹, Bahadır Feyzioğlu¹, Şerife Piringç², Mahmut Baykan¹

1. Department of Medical Microbiology, Selcuk University Meram Faculty of Medicine, Konya, Turkey
2. Department of Pediatrics, Subdivision of Chest Disease, Selcuk University Meram Faculty of Medicine, Turkey

✉ Corresponding author: Mehmet Özdemir, MD, Department of Medical Microbiology, Selcuk University Meram Medical Faculty, TR-42080, Konya, Turkey. Tel: 00903322236856; Fax: 00903322236181; email: mehmetozdem@yahoo.com

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Abstract

Sphingomonas paucimobilis, is a yellow-pigmented, aerobic, non fermentative, gram negative motile bacillus. *S. paucimobilis* which is widely found in nature and hospital environments rarely cause serious or life threatening infections. In this report, a case of hospital acquired bloodstream infection due to *S. paucimobilis* in a patient with Down syndrome who was on treatment for presumed pneumonia is presented.

A one year-old child patient who was a known case of Down syndrome and had previously experienced cardiac surgery was hospitalized and treated for pneumonia. On the 12th day of hospitalization, blood cultures were taken because of a high body temperature. One of the blood cultures was positive for gram-negative rods. After 48 hour of incubation, the sub-cultures on blood agar medium yielded pure growth of a yellow, non-fermentative, gram-negative, rod-shaped bacterium. The microorganism was positive for oxidase, and esculin hydrolysis, while negative for urea and nitrate reduction, citrate utilisation and motility. The isolate had been identified as *S. paucimobilis* by using Vitek 2 system. The antibiotic susceptibility test was also performed with the same system and the strain was found to be susceptible to piperacillin-tazobactam and other antibiotics. Treatment with intravenous piperacillin-tazobactam (150 mg/kg/day) was initiated. He responded well to the treatment and was discharged after 10 days. This case is reported to emphasize that *S. paucimobilis* should be kept in mind as a nosocomial infectious agent in patients with Down syndrome and immunosuppressive patients and the infections should be treated according to the sensitivity test results.

Key words: *Sphingomonas paucimobilis*, bacteriemia, hospital, infection, Down syndrome

INTRODUCTION

S. paucimobilis is a gram-negative, slightly motile, non-fermentative, oxidase positive opportunistic pathogen that rarely causes infections in humans, and forms yellow-pigmented S colony in blood agar (1). It is found in nature, notably in water and soil. It was first discovered as an agent in humans in 1977 and named *Pseudomonas paucimobilis*. It was renamed as

Sphingomonas paucimobilis in 1990 in accordance with phylogenetic data (1,2). Varieties of *Sphingomonas* are widely used in biotechnology due to their ability for synthesis and decomposition of macromolecules (3). In various studies, *S. paucimobilis* has been shown to be a causative agent of infection in immune-compromised patients, and in hospital acquired

postoperative endophthalmitis, septic shock, septic arthritis and osteomyelitis (1,4,5). In this report, the first case of *S. paucimobilis* identified in Turkey in a pediatric patient with Down syndrome is presented.

CASE REPORT

A twelve-month-old male patient who was brought to the pediatric outpatient clinic in March 2010 with complaints of high temperature, cough and wheezing was hospitalized with a presumptive diagnosis of pneumonia. Since the patient had exhibited stigmas of Down Syndrome at birth, a cytogenetic examination was conducted and he was diagnosed with 47 XY+21 chromosome and regular type Down Syndrome. The echocardiography conducted on the patient revealed enlargement of right heart cavities, Secundum Atrial Septal Defect (ASD), apical muscular Ventricular Septal Defect (VSD), Patent Ductus Arteriosus and pulmonary hypertension. After an angiography conducted in October 2009, it was decided that surgery be performed and ASD-VSD primary repair, Patent Foramen Ovale (PFO) ligation, and banding of Pulmonary artery were performed in November 2009. The patient, who was cognitively impaired in terms of neurological development stages and had a history of frequent infections, was hospitalized with the presumptive of bronchopneumonia. Chest x-ray of patient showed infiltration in left apex and by the auscultation bilateral rale and rhonchus were detected. Intra Venous antibiotic ceftriaxone was started. On the 12th day of the patient's hospitalization, blood cultures (BACTEC, Becton Dickinson, USA) were taken. On the 2nd day of incubation, passages were performed from the bottle where growth was identified to EMB and sheep blood agar (bioMerieux, France). Yellow-pigmented smooth colonies were seen on the blood agar. In gram staining of these colonies, thin gram negative bacilli were identified. The microorganism was positive for motility test, oxidase reaction, citrate reduction and esculin hydrolysis and negative for urea and nitrate reductions. The isolate was identified as *S. paucimobilis* by using Vitek 2 (bioMerieux, France) system. The microorganism was identified two different times from the the peripheral blood and no central venous catheter. Investigation of hospital environment specimens showed no *S. paucimobilis* growth. There was organism in the blood bottles during subculture. Antibiotic susceptibility test was also performed with Vitek 2 (bioMerieux, France) system and the strain was found to be susceptible to amikasin, gentamicin, levofloxacin, tigecycline, trimethoprim-sulfamethoxazol piperacillin-tazobactam, ampicillin, amoxicillin-clavunate, cefuroxime, cefuroxime-axetil, cefepime, imipenem,

meropenem and resistant to ceftriaxone, cefepime cephoxitin and cephtazidim. A 15-day treatment of intravenous piperacillin-tazobactam (150 mg/kg/day) was initiated. The fever of patient was disappeared. He responded well to the treatment and discharged.

DISCUSSION

S. paucimobilis is a yellow-pigmented, aerobic, gram negative bacillus that is motile with polar flagellation, non fermentative and is non spore forming (5-8). The bacteria is widely found in natural environment, especially in water and soil (3,5,8). At the same time, it was isolated from respirators, hemodialysis devices, distilled water containers and thermometer props in hospital environments (3,5,8,9). *S. paucimobilis* was shown to be an infective agent in cases of sepsis, peritonitis, catheter-related infections, osteomyelitis, and endophthalmitis (9,10).

A case of a postoperative endophthalmitis that was caused by *Sphingomonas paucimobilis* was reported by Seo et al in 2008 in Korea. This case demonstrates a delayed onset of postoperative endophthalmitis at 3 months after cataract extraction and posterior chamber intraocular lens implantation (1). Another case report was the 50-year-old male with end-stage renal disease who had been receiving continuous ambulatory peritoneal dialysis for peritonitis from Kocaeli, Turkey. It was an unusual pathogen for peritoneal dialysis (PD)-associated peritonitis (2). A rare case of nosocomial bacteriemia agent in bronchopneumonia patient who had a history hydrocephalus and experienced two ventriculoperitoneal shunt surgery was reported from Ankara, Turkey in 2008 (3). Reported another case is a patient with acute myeloid leukemia who developed *S. paucimobilis* bacteriemia complicated by septic shock just before receiving an autologous hematopoietic stem cell transplant at King Faisal Specialist Hospital and Research Centre in Riyadh (5). A nosocomial outbreak of *S. paucimobilis* bacteriemia in hemato/oncology unit was also reported in Ankara, Turkey in 2007. Four blood culture results were found positive for this microorganism (6).

S. paucimobilis is an opportunistic pathogen (8). It can be pathogen in immune-suppressed patients such as hematology and oncology patients and in patients with an underlying disease. In our case, too, since the patient suffers from Down syndrome and underlying diseases, he is susceptible to infection caused by this microorganism (3-6,10,11). On the basis of the relevant literature we have reviewed, we can say that this is the first case of *S. paucimobilis* in a patient with Down syndrome in Turkey and probably in the world.

There are cases in which this microorganism causes epidemics in hospitals (8). In such patients, the cause usually cannot be determined and are considered to have endogenous origin (3). However, *S. paucimobilis* may be found in tap water, distilled water, nebulizer, respirator, dialysis liquids and other equipment in hospitals and lead to nosocomial infection (8).

Among these species, there are also microorganisms like *S. mucosissima* and *S. adhesiva* which are not very important clinically. However, the best known and the main pathogen within this genus is *S. paucimobilis* (5,6). *S. paucimobilis* is rare in comparison to pseudomonas and other related species and has less pathogenic effect. This situation is accounted for by a lack of lipopolysaccharide in the cell wall and existence of glycosphingolipids instead (1,3). Antibiotic susceptibility of the microorganism varies from study to study (3,7). In our study, it was found to be susceptible to aminoglycosides, quinolones, trimethoprim-sulfamethoxazoles and cephalosporin apart from cephoxitin and ceftazidime.

In conclusion, although *S. paucimobilis* is a rare hospital infection, it should be kept in mind as a cause, by virtue of the fact that it is a part of the flora in natural environment, especially in patients with immunosuppressive diseases and patients who have other underlying diseases. Since antibiotic susceptibility also exhibits variation, patients should be treated in accordance with the antibiotic susceptibility results.

Conflict of Interest

The authors have declared that no conflict of interest exists.

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