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Case report

Sphingomonas paucimobilis presenting as acute phlebitis: A case report

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ABSTRACT

Sphingomonas paucimobilis is a strictly aerobic, non-spore-forming Gram-negative bacillus, ubiquitous bacterium, thought to be an opportunistic pathogen and is rarely reported in clinical settings. Here in, is the first case report of Acute Sphingomonas phlebitis secondary to intravenous (IV) drug use. We present the case of a 39-year-old male who initially presented with pain in his right upper extremity, fevers and chills of three week duration. He admitted to regularly injecting heroin in his distal right upper extremity with visible erythema, tenderness and streaking along the path of vein along the injection site. Radiographic studies including X-ray of the right arm, ultrasound and a subsequent MRI of the right arm were not significant for any osteomyelitis, deep venous thrombus, abscess, cellulitis, osteomyelitis, or pyomyositis. Blood culture grew Sphingomonas paucimobilis. Patient was initially started on vancomycin and piperacillin/tazobactam and subsequently switched to levofloxacin to complete a 14 day course. Patient admitted to using toilet water to mix his heroin which we suspect may have been the source of his bacteremia. Since it was first reported in 1979, a wide variety of community-acquired and hospital-acquired infections have been attributed to this Sphingomonas. It is ubiquitous to natural environment. We believe that due to its widespread habitat and ability to survive in stress conditions it could be a potential future threat in the era of increasing antimicrobial resistance globally. More research needs to be done on early identification, pathogenesis, treatment and eradication of the organism.

Introduction

Sphingomonas paucimobilis is a strictly aerobic, yellow-pigmented, glucose non-fermenting, oligotrophic, non-spore-forming Gram-negative bacillus with a single polar flagellum widely distributed in the natural environment [1]. Sphingomonas are ubiquitous bacteria that have been reported to be found in water (i.e. pipes, bathtubs, distilled water, and hemodialysis fluid) and soil [2]. Sphingomonas is thought to be an opportunistic pathogen and is rarely reported in clinical settings. To the best of our knowledge we present the first case report of acute Sphingomonas phlebitis secondary to intravenous (IV) drug use.

Case presentation

A 39-year-old male with past medical history of neurogenic bladder, epispadias with bladder exstrophy requiring a long-term indwelling catheter, and recurrent urinary tract infections presented to our hospital with a three week history of pain in his right upper extremity localized to the bicipital region that worsened with movement. He also reported experiencing intermittent fevers and chills for the past one week. The patient had a history of IV heroin and cocaine use and admitted to regularly injecting heroin in his distal right upper extremity. Physical exam demonstrated needle track marks on the right upper extremity along with erythema, tenderness, and streaking along the path of a vein along the injection site.

Vitals at presentation included fever (temperature: 104.9 °F), tachycardia (HR: 120 bpm), blood pressure: (107/67 mmHg), and normal respiratory rate (18 bpm). Notable labs on admission included normal white blood cell count of 7.88×10^9/L, mildly elevated creatinine 1.32 mg/dL, elevated liver enzymes (AST: 163 U/L, ALT: 249 U/L, ALP: 124 IU/L, T. Bili 0.5 mg/dL), normal lactic acid 1.8, normal creatinine kinase (25), mildly elevated CRP (3.89 mg/L), and a normal ESR (16 mm/h). Urinalysis showed 3+ esterase, nitrite positive, and packed WBC. A subsequent urine culture showed mixed growth and contaminants. A urine drug screen was positive for cocaine and opioids. With a history of IV drug use, the patient was also screened for hepatitis and HIV. Of note, hepatitis C antibody was positive with hepatitis C PCR resulting at 228,665 IU/ml; HIV-1 and HIV-2 antibody screening tests were negative.

The patient was initially started on vancomycin and piperacillin/tazobactam. An ultrasound duplex of the arm did not show any evidence of superficial or deep venous thrombus. An X-ray of the shoulder

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and elbow did not show any evidence of osteomyelitis. Subsequently the patient underwent a CT scan followed by an MRI of the right upper extremity that did not show any evidence of abscess, cellulitis, osteomyelitis, or pyomyositis. A chest X-ray and CT scan of the chest did not show any evidence of pulmonary embolus or pneumonia. An abdominal ultrasound did not show any evidence of liver heterogeneity to suggest cirrhosis or ascites. A renal ultrasound did not show evidence of pyelonephritis. The patient's blood culture grew Sphingomonas paucimobilis in both collected samples at 48 h which were sensitive to levofloxacin (MIC 0.5 mcg/mL) and trimethoprim/sulfamethoxazole (MIC < 20 mcg/mL). Based on patient's physical exam finding of, streaking along the injection site vein along with erythema and tenderness and no other identifiable source of infection, diagnosis of Sphingomonas phlebitis was made. His antimicrobials were initially narrowed to piperacillin/tazobactam, and subsequently to levofloxacin due to the resulting susceptibilities. Repeat blood cultures were negative and the patient was discharged to complete a 14 day course of antimicrobials. On further interrogation the patient admitted to using toilet water to mix his heroin which we suspect may have been the source of his bacteremia. He denied any history of needle sharing.

Discussion

Sphingomonas paucimobilis was first reported to cause infection in humans in 1979 when it was isolated from a pure culture specimen in a sailor who had developed a leg ulcer; and at this time it was known as Pseudomonas paucimobilis. It was renamed Sphingomonas paucimobilis in 1990 based on phylogenetic data [1]. Since then a wide variety of community-acquired and hospital-acquired infections including bacteremia, catheter-related infections, meningitis, peritonitis, osteomyelitis, endopthalmitis, septic arthritis, urinary tract infections, biliary tract infections, lung empyema, and pneumonia have been associated with Sphingomonas [2]. In a study by Toh et al. on S. Paucimobilis infection, about 50% of the patients had community acquired infections while 50% had hospital acquired infections [5]. Primary bacteremia and catheter related infections are most commonly described occurring in 35% and 33% of patients respectively in another study [1]. Risk factors that have been reported include malignancy, diabetes, alcoholism, immunosuppressed states, liver cirrhosis, ESRD, COPD, and catheter-related infections [1]. Mean age has been reported to be 48 years with range from 5 months to 87 years [1]. Sphingomonadaceae are reported to be ubiquitous in the natural environment [1]. They have even been isolated from ultrapure distilled water systems, space shuttles, dental irrigation systems, hemodialysis-associated fluid systems and hospital water systems [2,3].

Due to the widespread habitat of sphingomonadaceae and their ability to survive in stress conditions they could play a pivotal yet under-recognized role as reservoirs of antimicrobial resistance in drinking water [2]. The widespread prevalence and ability to cope with environmental stress could be due to various factors. S. paucimobilis is an oligotrophic bacterium meaning that it has the ability to survive in low-nutrient environments [3]. It also has the ability to form biofilms in plumbing systems thus causing biological accumulation and spreading in industrial and drinking water systems. Moreover it can pass through 0.2 um filters that are traditionally used for terminal sterilization of several medicinal products [3]. The exact pathogenesis remains unknown, though the presence of pili, phosphpholipase C, protease, and a flagella gene are all thought to play a role [6]. The organism lacks the lipopolysaccharide (exotoxin) component typically found in the outer membrane of gram-negative bacterial cell walls. The lack of endotoxin activity may explain the lack of virulence and the favorable prognosis of S. paucimobilis infections [1].

Diagnosis is based on isolating the organism from culturing in appropriate media. No controlled trials exist regarding therapy. Conflicting data exists regarding antimicrobial susceptibility: initial isolates were reported to be susceptible to tetracycline, tobramycin, kanamycin, gentamicin, sulfamethoxozole and carbenicillin, but resistant to ampicillin, colistin, cephalothin and streptomycin. Other authors have reported their Sphingomonas isolates to be resistant to beta-lactam's. Vaz-Moreira isolated 86 sphingomonadaceae isolates from drinking water and reported them to be intrinsically resistant to colistin. They reported more than 50% of strains were resistant to beta-lactam's, followed by fluoroquinolone (25.6%) and sulfonamides (20.9%). An interesting finding in their study was that Sphingomonas had the widest range of resistance phenotypes, comprising resistance to 18 of 19 antimicrobials tested in their study [2]. These differing results indicate that the appropriate antimicrobial therapy may need to be chosen on an individualized basis. In our case, the bacteria was sensitive to gentamicin, levofloxacin, meropenem, tobramycin, trimethoprim/sulfamethoxazole; it was resistant to piperacillin/tazobactam. Aminoglycosides plus third generation cephalosporins or cabepenems have been reported as suitable initial treatment regimens, along with identification and removal of the source if possible. Fluoroquinolones have been reported to be most efficacious antimicrobial agents [1]. Ampicillin/sulbactam, piperacillin/tazobactam and imipenem have also been reported to be effective [1]. Survival has been reported to be favorable, reportedly as high as 95%, despite initial usage of inappropriate antimicrobials [5]. The mortality rate is about 6% [4]. Despite this low virulence, early isolation and treatment is essential as this pathogen can cause life-threatening septic shock, particularly in immunocompromised hosts [3].

Conclusion

In conclusion, Sphingomonas can cause a variety of community-acquired as well as nosocomial infections in both immunocompetent and immunocompromised individuals. It needs to be recognized as an important infectious pathogen that should be noted on the differential diagnoses of patients with bacteremia of unknown etiology. We believe that it is likely under-reported; however, more research needs to be done on pathogenesis, early identification and treatment of the organism as Sphingomonas can lead to septic shock if inappropriately treated. It seems reasonable to conclude that in the era of increasing anti-microbial resistance globally, more awareness should be raised about this bacterium since it could be a potential future threat.

Author contributions

SW, AM, NH reviewed the case and literature and drafted the manuscript; TL reviewed the literature, made critical revisions related to the content of the article and approved the final version of the article to be published. Informed patient consent was obtained for publication purposes. All authors read and approved the final manuscript for submission.

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Ethics approval

Does not apply.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Availability of data and material

Data sharing is not applicable to this article as no data sets were generated or analysed during the current study.

Competing interests

No conflicting interest related to the manuscript submitted for publication.

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