Case report

A possible Sphingomonas paucimobilis endocarditis in a haemodialysed patient: A case report

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Abstract

Sphingomonas paucimobilis is widely distributed in the environment and causes occasional human infections, most often in immunocompromised individuals. Reports of unusual invasive and severe infections include septic arthritis, osteomyelitis, respiratory tract infections in patients with cystic fibrosis and necrotising soft tissue infections. There are only few reported cases of endocarditis in the international literature. There are no published cases in Sri Lanka.

We describe a possible case of Sphingomonas paucimobilis endocarditis. A 58 year old male patient presented with reduced responsiveness following haemodialysis. All three blood cultures taken on admission were positive for Sphingomonas paucimobilis. His echocardiography could not exclude endocarditis. He was treated with appropriate antibiotics but unfortunately expired due to septic shock following aspiration pneumonia. This case demonstrates that organisms of low virulence could cause endocarditis in patients with end stage renal disease. It also indicates that such patients are relatively asymptomatic or have few symptoms compared to patients with normal immunity.

Keywords: Sphingomonas paucimobilis, endocarditis, Sri Lanka

Introduction

Sphingomonas paucimobilis is widely distributed in soil and water, including water sources in the hospital environment. It is an occasional human pathogen. This organism was formerly known as Pseudomonas paucimobilis. Most Sphingomonas infections are hospital acquired and typically occur in immunocompromised individuals.¹ Although cases have been reported internationally, there is no published data on Sphingomonas paucimobilis bacteraemia nor on endocarditis in Sri

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Lanka. We present a possible case of *Sphingomonas paucimobilis* endocarditis in a 58 year old male patient.

Case report

A 58 year old male on regular haemodialysis was admitted with sudden onset of reduced responsiveness with GCS 3/15 after haemodialysis (day 1). His blood pressure was 180/100 mmHg with evidence of ankle oedema. Prior to dialysis, the patient was fully conscious with GCS 15/15 but had fever on and off for 3 days. Vancomycin IV 1g stat and 500mg after each dialysis was administered to the patient as a blood culture from the vascular catheter line had grown methicillin sensitive *Staphylococcus aureus* (MSSA) three days prior to admission.

He was a known patient with type 2 diabetes mellitus, end stage chronic kidney disease (CKD), hypertension, ischaemic heart disease, antral gastritis, chronic calculous cholecystitis, pulmonary tuberculosis in 2019 and COVID-19 infection in 2021.

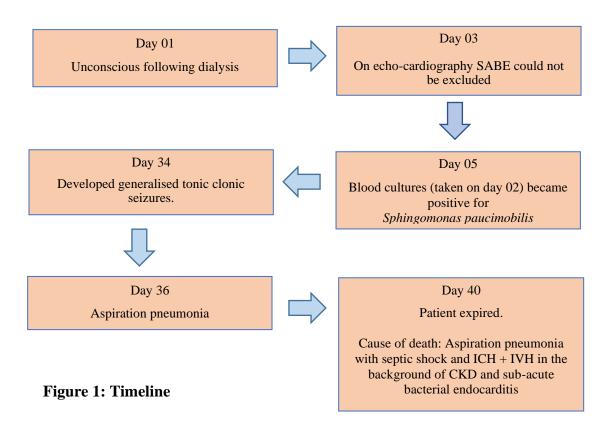
All three peripheral blood cultures sent on day two of admission were positive for Gram positive bacilli and the isolate was sent to two centres for further identification. It was identified as *Sphingomonas paucimobilis* by the two centres using BD Phoneix® and Vitek2® automated identification systems. The isolate was sensitive (detected using automated system Vitek2®) to piperacillin-tazobactam (MIC 16 mg/L), cefepime (MIC 4 mg/L), imipenem (MIC 1 mg/L), tigecycline (MIC <= 0.5 mg/L) and co-trimoxazole (MIC 20 mg/L). The isolate demonstrated intermediate sensitivity to ceftriaxone (MIC 16 mg/L), cefoperazone-sulbactam (MIC 32 mg/L), meropenem (MIC 8 mg/L), gentamicin (MIC 8 mg/L) and ciprofloxacin (MIC 2 mg/L) but was resistant to amikacin (MIC >= 64 mg/L). He was started on piperacillin-tazobactam renal adjusted dose. A repeat peripheral blood culture taken on day nine again signalled positive and had the same isolate. Subsequent blood cultures were sterile. Echocardiography done on day three showed an ejection fraction of 45%-50%, Grade II MR with annular calcifications with myxomatous material, and hence subacute bacterial endocarditis (SABE) could not be excluded. Due to haemodynamic instability, the patient was unable to undergo trans-oesophageal echocardiography.

Non contrast computerised tomography (NCCT) of the brain was done on day one which showed right fronto-temporal intracranial haemorrhages (ICH) with significant peri-lesion oedema and intraventricular haemorrhages (IVH) with midline shift. Follow up NCCT on the brain done on day 36 did not show any expansion. The patient was managed conservatively as the neurosurgical opinion was that the prognosis was poor.

Piperacillin-tazobactam 2.25g 6 hourly was given for 17 days. The patient developed diarrhoea on day 17 and piperacillin-tazobactam was omitted as antibiotic induced diarrhoea was suspected. The patient was then started on IV ceftriaxone 2g daily which was given for another 17 days. IV vancomycin 500 mg after each dialysis was given for 23 days and IV metronidazole 500mg tds was given for 4 days (started on day 36). He was dialysed twice a week throughout the hospital stay.

The patient developed generalized tonic-clonic seizures on day 34 with vomiting. He became drowsy again which was considered to be due to either postictal drowsiness or scar epilepsy. He

developed pneumonia following seizures and was diagnosed to have aspiration pneumonia. The patient expired on day 40 and cause of death was given as "Aspiration pneumonia with septic shock and ICH + IVH in the background of CKD and subacute bacterial endocarditis".



Discussion

Sphingomonas paucimobilis is a polymorphic Gram-negative strict aerobic bacillus, which is a weakly oxidase positive, catalase positive, and yellow pigment producing organism that grows on blood agar and does not grow on MacConkey agar. Despite the presence of a single polar flagellum, only a low percentage of cells are actively motile, and motility can be difficult to demonstrate in the laboratory (thus the name paucimobilis). There are no reports of gram variability observed in the organism, but all the isolates appeared gram positive, although identified as Sphingomonas paucimobilis at two different centres. Two centres were used, as MIC values for all antibiotics were not available from the first centre. The primary ABST suggested that the isolate was Gram negative because of resistance to vancomycin. The second isolate on day 9 showed the same Gram positive appearance. Since the isolates were repeatedly stained with the same set of stains, it is unlikely to be an error of the staining. A literature search on Gram stain of S. paucimobilis did not provide an explanation for this anomaly.

The 3 samples for blood culture taken on day 2 after admission were positive for *Sphingomonas* paucimobilis satisfying the following diagnostic criterion for infective endocarditis: All 3 blood

cultures taken on day 2 of admission positive for the same organism with first and last sample drawn at least 1 h apart. However, the valvular lesion could not be confirmed by TEE.

Though *S. paucimobilis* lacks endotoxins, patients can end up in septic shock. Random and unusual invasive and severe infections including septic arthritis and osteomyelitis, respiratory tract infections in patients with cystic fibrosis and necrotising soft tissue infections have been reported. ^{2,3} Diabetes mellitus and alcoholism appear to be risk factors for community-acquired *S. paucimobilis* infections, including bacteraemia.⁴ Malignancy, end-stage renal disease, diabetes mellitus, and use of immunosuppressive drugs are the most common risk factors,⁵ and our patient had end-stage renal disease and diabetes mellitus.

S. paucimobilis is not included as a cause of endocarditis in the international literature. A case report of S. paucimobilis endocarditis in an immune-competent patient in the United States without any comorbidities or risk factors, and successfully managed with appropriate antibiotics for 6 weeks suggested the importance of considering S. paucimobilis in the differential diagnosis of infective endocarditis, even in patients with normal immunity. S. paucimobilis tricuspid valve endocarditis in an intravenous drug user who responded rapidly to meropenem and levofloxacin has also been reported in the USA. The importance of opportunistic gram negatives in clinical settings has been shown by a case report of IE in a patient on immunosuppressive therapy for six years who recovered with meropenem for 6 weeks. A fatal multi-drug resistant S. paucimobilis prosthetic valve endocarditis case has been reported in Indonesia.

Our patient also received antibiotics for 34 days (until his death) and expired because of septic shock resulting from aspiration pneumonia. A repeat echocardiogram was not done so we are unable to comment on echocardiographic improvement. This patient had many complications, and relatives were informed about the poor outcome from the beginning. A pathological post-mortem was not carried out.

The resistance rates reported in USA for *S. paucimobilis* isolates are usually 0% for amikacin, ciprofloxacin, and imipenem, 5% for cefepime and 10% for ceftazidime ⁷ It was difficult to obtain an accurate antibiotic history of this patient. However, it is unlikely that the patient was prescribed imipenem, as patients with CKD do not receive imipenem due to nephrotoxicity. Cefepime is not available in Sri Lankan hospitals. Amikacin is sometimes given to CKD patients when lines are colonised with multidrug resistant Gram-negative bacilli. Ciprofloxacin is used widely and intermediate sensitivity of *S. paucimobilis* to ciprofloxacin is unexpected.

The three blood cultures taken on day 2 and the blood culture taken on day 9 were positive and all isolates were identified as *B. paucimobilis* and had the same antibiotic sensitivity profile.

There is no published data or case reports on *S. paucimobilis* endocarditis in Sri Lanka. Availability of automated identification for identification of Gram negative bacilli made it possible to identify the isolate from this patient.

Take home message.

Final identification to species level of isolates from blood cultures or sterile sites is very important. Patients with end stage renal disease are likely to get endocarditis with organisms of low virulence and may not show many symptoms unlike patients with normal immunity. Fever in these patients require full investigation with blood cultures unless there is an obvious alternate cause. *S. paucimobilis* is of increasing importance and should be considered as an aetiological agent for infective endocarditis and sepsis in an immunocompromised patient.

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