



AORTIC BIOPROSTHETIC VALVE INFECTIVE ENDOCARDITIS DUE TO BURKHOLDERIA CEPACIA COMPLEX BACTREMIA IN AN IMMUNOCOMPETENT INDIVIDUAL WITH SUCCESSFUL TREATMENT – A CASE REPORT

Jagadeesh Chandrasekaran, Jovitha Preethi Gregory, Mathi Manoj Kumar R*, P Senthur Nambi, and Ram Gopalkrishnan

Department of Internal Medicine, Apollo Hospitals, Greams Lane, Chennai-600006, Tamil Nadu, India

ARTICLE INFO

Article History:

Received 10th October, 2018

Received in revised form 2nd

November, 2018

Accepted 26th December, 2018

Published online 28th January, 2019

Key words:

Burkholderia Cepacia Complex, Prosthetic Valve Endocarditis

ABSTRACT

Prosthetic valve endocarditis is a direful complication of cardiac valve replacement and is associated with high mortality rates. There has been a recent change in the microbial spectrum of pathogens. One among them is Burkholderia cepacia complex (BCC), which is rare but serious, leading to valve dysfunction and heart failure thereby resulting in significant morbidity and mortality. BCC is intrinsically resistant to multiple antibiotics which lead to frequent treatment failures. BCC is known to cause serious infections in patients with cystic fibrosis¹, chronic granulomatous disease, malignancies, (Intravenous drug) IVD abuse, and is also transmitted nosocomially. Here, we report a case of successful treatment of BCC prosthetic valve endocarditis with splenic hypodense lesions, in an immunocompetent individual, which was detected in the second (Trans Esophageal Echocardiogram)TEE. To the best of our knowledge, there are only very few reported cases of infective endocarditis(IE) due to BCC in the medical literature². The need for a better awareness among clinicians on the recent trend of changing pathogens implicated in IE, and a strong clinical suspicion for the same, is essential for accurate management.

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INTRODUCTION

BCC, an aerobic Gram-negative bacillus and an emerging bacterial pathogen, is isolated mostly from patients who have compromised immunity, or are acquired nosocomially. BCC is a rare cause of infective endocarditis, with very few reported cases in the medical literature². It is the 4th commonest pathogenic NFGNB (non-fermenter gram negative bacilli) identified worldwide, the initial 3 being Pseudomonas aeruginosa, Acinetobacter baumannii and Stenotrophomonas maltophilia³. It is an emerging pathogen of clinical importance, due to its difficulty in laboratory isolation and intrinsic extensive antibiotic resistance. BCC recently is found to have been reported even in patients who are not known to have any predisposing factors⁴. Initially being introduced in the field of agriculture due to its fungicidal properties as a biopesticide in 1950's, it was known to produce onion rot in 1959, and was found to be associated with severe respiratory tract infection in patients with cystic fibrosis in 1980's. The first patient-to-patient spread was eventually reported in 1990's,⁵.

Case Report

A 66 year old gentleman, known hypertensive, with a history

of curative right hemicolectomy for carcinoma caecum in 2008, curative total left nephrectomy for renal cell carcinoma and cholecystectomy in 2011, hernioplasty in 2014, aortic valve replacement for severe calcific aortic stenosis in 2016 with a bioprosthetic valve, and right hip replacement for right acetabular comminuted fracture with sciatic nerve palsy following an RTA in May 2018, presented with a ten day duration of low grade fever, dry cough, loss of appetite and burning micturition. He was a non-smoker, non alcoholic, with no history of intravenous drug abuse. On arrival, he was drowsy, febrile, tachycardic, and required inotropic support. Physical examination showed pallor and a systolic murmur in the aortic area. Baseline blood tests revealed anaemia, severe hyponatremia requiring 3% hypertonic saline infusion, hyperkalaemia and mildly elevated liver enzymes. Trans thoracic echo showed an aortic bioprosthetic valve with Aortic Valve Mean Pressure Gradient of 17 mmHg, mild PAH and absent vegetations. Suspecting urosepsis, he was initiated empirically on IV Cefoperazone- sulbactam, after sending samples for urine culture and 2 sets of blood cultures. USG abdomen showed a splenomegaly (14.3 cm). Urine cultures grew CR (Carbapenemase Resistant) E coli, for which he was started on Injection Colistin. Two aerobic sets of blood cultures, sampled before the commencement of antimicrobial therapy, yielded growth of Burkholderia cepacia complex, which was highly drug resistant (Figure 2). Sensitivity results-directed therapy consisting of Injection Ceftazidime was

*Corresponding author: Mathi Manoj Kumar R

Department of Internal Medicine, Apollo Hospitals, Greams Lane, Chennai-600006, Tamil Nadu, India

commenced. Despite being on sensitive antibiotics, the overall clinical condition of the patient did not improve. HIV was negative and he was non-diabetic. Further work-up detected secondary HLH (Hemophagocytic Lymphohistiocytosis) due to the ongoing sepsis. In total, four blood cultures drawn on days 1, 11, 18 and 23, were all positive for the same organism but with varying sensitivity pattern. A CECT abdomen was done on the 7th day of admission, which showed splenomegaly (17 cm) and multiple ill-defined hypodense lesions, which was thought to be either infarcts or abscesses (Figure 1). A TEE was done on the 10th day of admission to rule out a bioprosthetic valve infective endocarditis, which was negative for vegetations. He also developed right leg cellulitis during the hospital stay, which was managed conservatively.



Figure 1 Splenic hypodense lesions in the CECT abdomen

CULTURE AND SENSITIVITY [BLOOD] : (Culture)	
Gram Stain	-
Organism Name	<u>Burkholderia cepacia complex</u>
Methodology Name	Kirby Bauer
Growth Observed	Yes
Antibiotics Name	Results
Gentamicin	Resistant
Co-Trimoxazole	Sensitive
Amikacin	Resistant
Ceftazidime	Sensitive
Ciprofloxacin	Sensitive
Cefepime	Sensitive
Imipenem	Resistant
Meropenem	Intermediate
Cefoperazone+Sulbactam	Sensitive
Piperacillin + tazobactam	Sensitive
Report Status: Final	

Figure 2 Initial positive blood culture report

CULTURE AND SENSITIVITY [BLOOD] : (Culture)	
Gram Stain	-
Identification Method	Vitek 2 compact
Organism Name	<u>Burkholderia cepacia complex</u>
Methodology Name	Kirby Bauer
Growth Observed	Yes
Antibiotics Name	Results
Gentamicin	Resistant
Co-Trimoxazole	Sensitive
Amikacin	Resistant
Ceftazidime	Resistant
Ciprofloxacin	Resistant
Cefepime	Resistant
Imipenem	Resistant
Meropenem	Resistant
Cefoperazone+Sulbactam	Resistant
Piperacillin + tazobactam	Resistant
Doripenem	Resistant
Aztreonam	Resistant

Minocycline MIC: 8 ug/ml (Intermediate)
Report Status: Final
* END OF REPORT *

Figure 3 and 4 Final positive blood culture reports

His general condition started deteriorating over the next few days with spiking fever, tachycardia and persisting drowsiness. In view of the negative TEE, multiple splenic abscesses were suspected as the cause for the persisting Burkholderia cepacia bacteraemia, as suggested by the CECT W/A, and hence an open splenectomy was done on the 18th day of admission.

Splenic lesions were multiple, which rendered the less invasive percutaneous CT guided drainage a difficult approach for source reduction. New sets of blood cultures showed persistent BCC bacteraemia, sensitive to Cotrimoxazole and Piperacillin/tazobactam. Sensitivity results-directed therapy consisting of Cotrimoxazole, and Minocycline (with an MIC less than 4) was commenced, in view of an allergic reaction to Piperacillin- tazobactam on initiation. Post Splenectomy, he remained intubated for a few days, and was on Volume control mode of ventilation. He had intra-operative massive blood loss upto 4 Litres, due to the multiple intra abdominal adhesions, for which he received PRBC transfusions. Splenic pus culture was sterile, and HPE showed multi-focal infarcts with foci of necrosis, suggesting embolic infarctions of the spleen.

On 36th day of admission, he had chest pain with ECG changes and Trop I positivity. The screening TTE showed a normal EF. A TEE was done for the second time in the background of a strong clinical suspicion, which finally revealed a 16 mm vegetation attached to the aortic bioprosthetic valve, with 2+AR and moderate PAH. A diagnosis of bioprosthetic aortic valve infective endocarditis was made according to the modified Dukes criteria. It was decided to go ahead with conservative management for the same.

CT cardiac angiography with gated CTA of thoracic aorta was done on 40th day of admission, which ruled out further complications like aortic root abscess. Aortic valve showed insignificant pannus, but did not show any vegetation. MRI pelvis was also done in order to rule out a right hip implant infection, which showed intra abdominal collection. The intra abdominal collection was drained, and was cultured, which grew Multi drug resistant (MDR) Klebsiella sensitive to colistin. A TTE was repeated on the 50th day of admission, which did not show any vegetation over the aortic bioprosthetic valve, and hence conservative care was continued, after considering the surgical morbidities and an increased risk for valve surgery. A repeat blood culture was done, prior to discharge, which was sterile. He was discharged on the 59th day of admission, with oral Minocycline and Co-trimoxazole, both for a period of 3 months. Patient was discharged from the hospital with appropriate treatment, and did very well at OPD Follow-up, and completed the course oral antibiotics.

DISCUSSION

BCC organisms are a group of ten closely-related species of aerobic environmental Gram-negative bacteria.³

BCC organisms are extremely rare causes of endocarditis, with very few cases reported till date in the English Medical literature. In a report at JMM, Seven of these infections occurred on native valves, and in six instances, patients were intravenous drug abusers¹.

Because Burkholderia is a microbe uncommonly found on human skin, this infection can only occur if the person involved was somehow exposed to water contaminated by this organism. Advances made possible by technologies in medical science, such as the use of intravascular catheters in the management of patients in intensive care units, have been shown to be one of the many ways that these newly recognized organisms enters into the human body⁵.

BCC is resistant to multiple antibiotics which leads to frequent treatment failures⁶. Therapy for this organism must be tailored on the basis of the culture and sensitivity result⁵.

TEE is more sensitive than transthoracic echocardiography (sensitivity 82-96% versus 17-36%) in detecting vegetations as small as 1-2 mm in size as well. The literature about the optimal time interval for repetition of the examinations is scanty. Repeated TEE-examinations has been recommended as a diagnostic strategy in suspected endocarditis, with a suggested time interval of 7-10 days for repeating echocardiograms. Vegetations on prosthetic valves are more difficult to detect by TTE than those involving native valves, and therefore TEE should always be used if the diagnosis of prosthetic endocarditis is suspected.

CONCLUSION

The above patient had a persistent BCC bacteraemia identified early in the initial blood cultures, with unidentified source, which was evident, only on the second repeated TEE. He had to undergo an open splenectomy; he had a prolonged hospital stay in view of his poor clinical condition and for appropriate treatment. He had three foreign bodies namely, bioprosthetic aortic valve, abdominal wall mesh, and the right hip plate, any of which could have been the source of the persistent bacteraemia. He had a history of two treated malignancies in the past, and multiple contacts with health care facilities and invasive arterial and venous catheters, which might well have played an important role in the colonisation and infection of this patient by this bacterium.

In summary, this report is unique in the literature, as it describes a case of successful treatment of prosthetic valve infective endocarditis due to BCC along with splenic infarcts, and illustrates the need for a repeated TEE⁷, which was done twice in our case, in the presence of a strong clinical suspicion of infective endocarditis due to this uncommon pathogen.

Conflicts of Interest: None

How to cite this article:

Jagadeesh Chandrasekaran *et al* (2019) 'Aortic Bioprosthetic Valve Infective Endocarditis Due to Burkholderia Cepacia Complex Bactremia in an Immunocompetent Individual With Successful Treatment – A Case Report', *International Journal of Current Advanced Research*, 08(01), pp. 17135-17137. DOI: <http://dx.doi.org/10.24327/ijcar.2019.17137.3198>

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