

***Kytococcus Schroeteri* Prosthetic Valve Endocarditis Complicated by Aortic Pseudoaneurysm: A Rare Complication from A Rare Agent**

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ABSTRACT

Kytococcus schroeteri is a rarely described slow-growing gram-positive coccus, with recent association with Infective Endocarditis cases. A 29-year-old male with prior history of rheumatic valvular heart disease and HIV infection, previously submitted to two cardiac surgeries, the last one 12 years ago with mechanical mitral and aortic valve implantation plus tricuspid annuloplasty was evacuated from Cape Verde with congestive heart failure and no features of infection. The diagnostic work-up revealed an aortic paravalvular leak and a large pseudoaneurysm as well as a severe tricuspid regurgitation. The patient underwent surgery with Bentall procedure and a mechanical tricuspid valve implantation. The microbiologic exam of the removed aortic valve using 16sRNA sequencing showed a *Kytococcus schroeteri*, revealing its association with a severe complicated case of prosthetic valve endocarditis.

Keywords: *Kytococcus Schroeteri*, Prosthetic Valve Endocarditis, Aortic Pseudoaneurysm

Introduction

Kytococcus schroeteri is a gram-positive coccus (Fernández *et al.*, 2021), a skin commensal bacteria first distinguished from *Micrococcus* in 1995. Eight years later it was associated with cases of infective endocarditis (IE), with very few cases reported so far (Al Tamtami *et al.*, 2019). These agent's colonies show spherical cells, displayed in pairs, in tetrads or, occasionally, in clusters (Mnif *et al.*, 2006). Also, differently from *Micrococcus* spp., *Kytococcus* spp. are often penicillin and oxacillin resistant and have arginine dihydrolase activity. It's also resistant to cephalosporins, quinolones and macrolides (Fernández *et al.*, 2021; Al Tamtami *et al.*, 2019). Its detection is not possible with the usual microbiologic cultural exams as the blood cultures, needing molecular methods, the main one 16s rRNA sequencing. Aortic pseudoaneurysm is a rare and severe complication of IE typically associated with bad prognostic (Wang *et al.*, 2015). The

damage caused by prosthetic aortic valve endocarditis can lead to destruction or perforation of the leaflet, ring abscess, perivalvular regurgitation and less commonly result in damage to mitral- aortic intervalvular fibrosa (MAIVF) leading to an aortic pseudoaneurysm (Wang *et al.*, 2015). In a series of 818 cases of IE, it was reported a 2% rate of pseudoaneurysm of the MAIVF (Afridi *et al.*, 1995). In this report it is described and discussed the first case of *Kytococcus schroeteri* prosthetic endocarditis complicated with pseudoaneurysm.

Clinical Case Report

A 29-year-old male with rheumatic valvular heart disease was evacuated from Cape Verde with NYHA (New York Heart Association) class III heart failure (HF) and suspected aortic paravalvular leak.

The patient was previously submitted to an aortic homograft implantation plus mitral valvuloplasty 21 years ago due to aortic and mitral significant regurgitations in the context of rheumatic carditis and a re-submitted to a surgical procedure with double mechanical mitral (St Jude) and aortic (St Jude 19) valve implantation plus tricuspid annuloplasty (CE 32) 12 years ago, due to homograft dysfunction and severe mitral and tricuspid regurgitation. The patient also has HIV infection but was compliant with the anti-retroviral medication and has no detectable viral load.

At admission, the patient showed signs of acute decompensated HF, with pulmonary crepitations, ascites and peripheral edema. Blood tests revealed a hemolytic anemia (hemoglobin 10.3g/dL, hyperbilirubinemia, LDH 1200 U/L, haptoglobin <0.07 and schistocytes in blood smear) due to mechanic valvular trauma, no leukocytosis or C-reactive protein elevation, normal renal function and elevated natriuretic peptide. Blood culture tests were negative.

Transthoracic echocardiogram (TTE) and Transesophageal echocardiogram (TEE) were performed (Fig. 1-2) and showed left ventricle hypertrophy, bi-atrial enlargement, mitral valve prosthesis with adequate gradients and no significant regurgitation, aortic prosthesis with 32mmHg peak gradient, valve aortic moderate to severe peri-valvular leak and a very large pseudoaneurysm that surrounded the aortic valve, tricuspid ring in the adequate position with severe regurgitation, dilated right ventricle and estimated pulmonary artery systolic pressure of 82mmHg.

A computed tomography (CT) scan was performed (Fig. 3) and confirmed the presence of a very large pseudoaneurysm with axial diameters 107x69 mm, with communication with the left ventricle outflow tract, immediately inferior to the aortic valvular plane in posterior topography. The CT-scan also excluded the presence of aortic dissection.

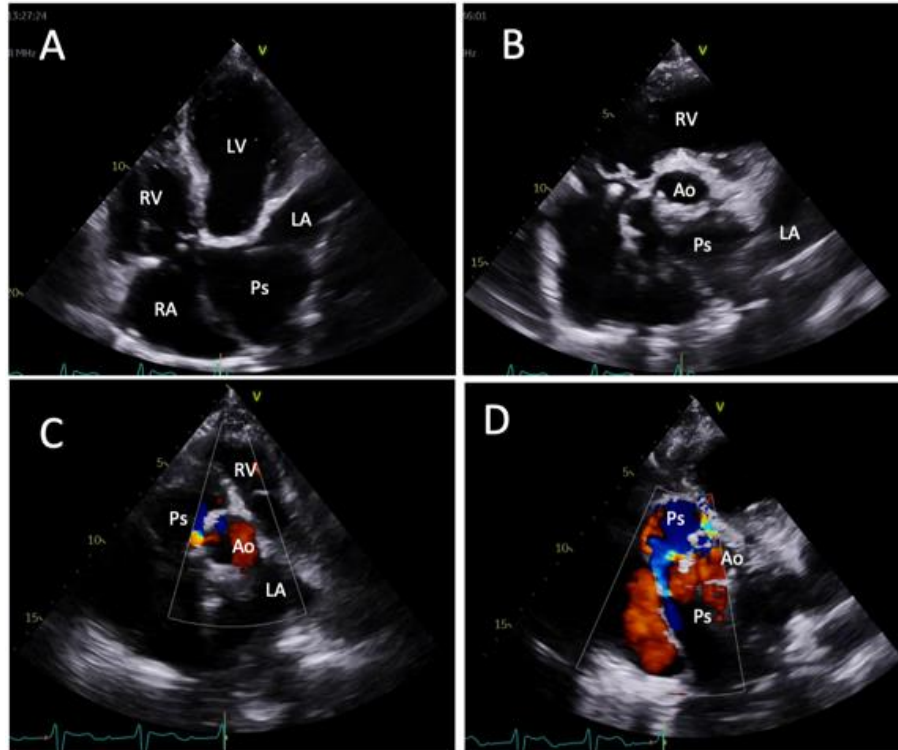


Figure 1: Transthoracic echocardiogram showing a structure near the left ventricle outflow tract (LVOT) and left atrium in apical 4-chamber (A), localized posteriorly of the aortic valve in parasternal short axis (B) with communication with the LVOT visualized with color doppler (C and D). (LV – left ventricle; RV – right ventricle; LA – left atrium; RA – right atrium; Ao – aorta; Ps – pseudoaneurysm).

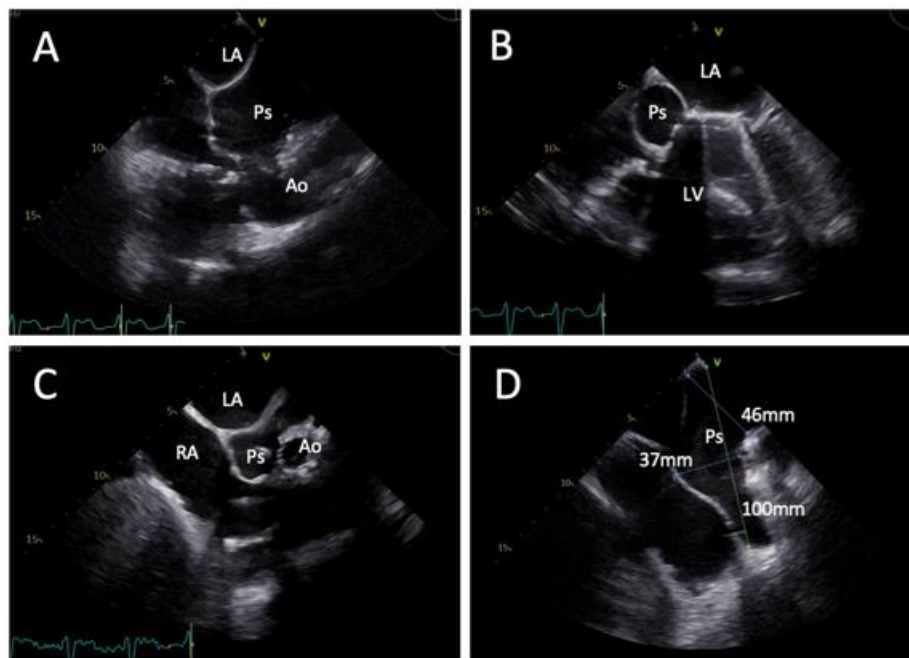


Figure 2: Transesophageal echocardiogram showing the pseudoaneurysm posteriorly to the LVOT, aortic valve and ascending aorta (A, B and C), maximum dimensions 100x46x37mm (D). (LV – left ventricle; RV – right ventricle; LA – left atrium; RA – right atrium; Ao – aorta; Ps – pseudoaneurysm).

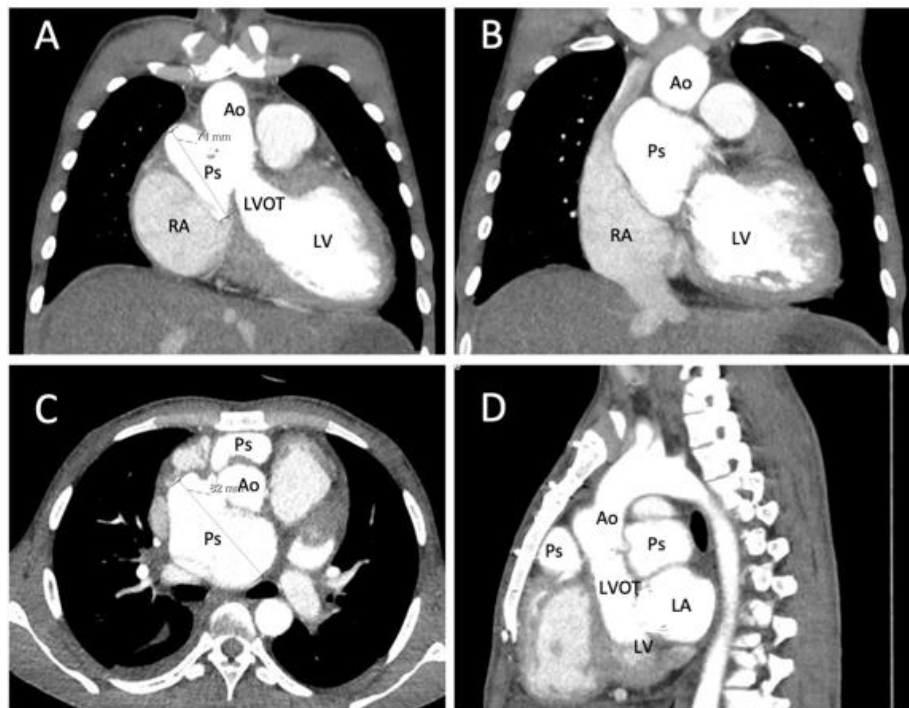


Figure 3: Computed tomography scan confirming the aortic pseudoaneurysm involving part of the LVOT, aortic valve, aortic root and ascending aorta in coronal (A and B), axial (C) and sagittal (D) planes. (LV – left ventricle; LVOT – left ventricle outflow tract; LA – left atrium; RA – right atrium; Ao – aorta; Ps – pseudoaneurysm).

Antibiotic therapy was not started before surgery because of the normal acute phase parameters and lack of microbiological agent in the blood cultures. Through median sternotomy and under extracorporeal circulation, aortotomy was performed, the pseudoaneurysm and prosthetic aortic valve were excised, followed by a Bentall procedure (composite of graft replacement of the aortic valve, aortic root and ascending aorta with re-implantation of the coronary arteries into the graft) with an aortic mechanical valve, size 21mm. The mitral prosthesis was not involved in the infective process and was preserved. Through a right atriotomy, the tricuspid ring was excised and a mechanical tricuspid valve size 29mm was implanted. The surgery was performed with TEE support, that confirmed the absence of residual perivalvular leak. The surgery lasted 5 hours and 45 minutes (345 minutes), with an extracorporeal circulation time of 255 minutes and an aortic cross-clamp time of 189 minutes. The patient was admitted in Cardiac Surgery Intensive Care Unit with only mild inotropic support, he was extubated at 6 hours after the procedure, there was no need for transfusional support or renal replacement therapy.

The excised aortic prosthesis microbiologic analysis revealed growth of *Kytococcus schroeteri* (through PCR 16s rRNA sequencing). 3 days after surgery the patient started antibiotherapy with vancomycin (six weeks) plus rifampicin (five weeks) plus gentamicin (two weeks), with good clinical response. Post-operative TTE (Fig. 4) showed normal function of the aortic, mitral and tricuspid mechanical

valves, without peri-valvular leaks, good left ventricular systolic function, reasonable right ventricular systolic function, bi-atrial enlargement and mild posterior pericardial effusion. The patient was discharge under medication with acenocumarol, bisoprolol 2.5mg id, ramipril 2.5mg id, furosemide 40mg bid, spironolactone 100mg id and tenofovir/emtricitabine/dolutegravir.

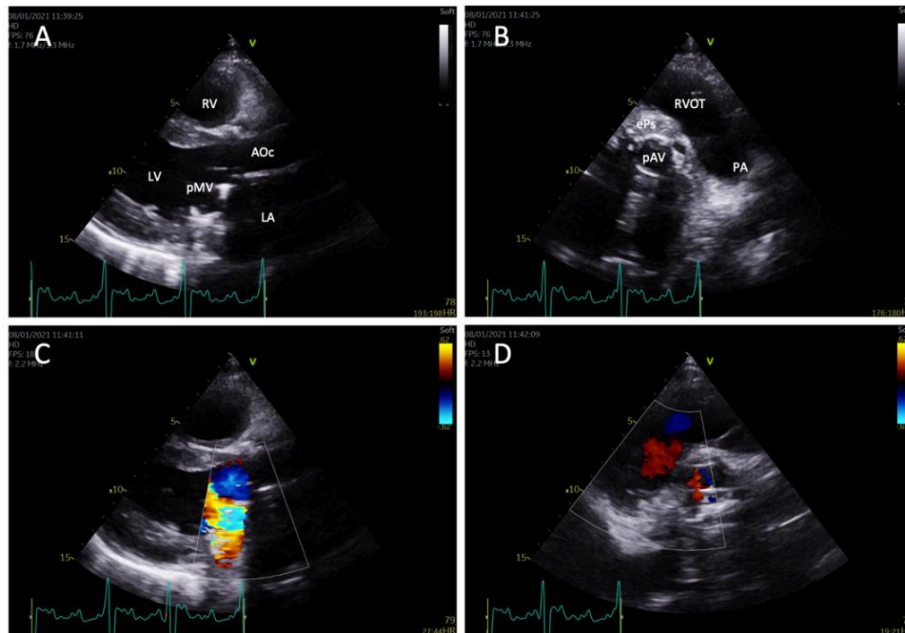


Figure 4: Post-operative Transthoracic echocardiogram showing adequate position of the aortic conduit with good exclusion of the pseudoaneurysm and a well-functioning mechanical aortic valve, without peri-prosthetic leak (RV – right ventricle; LV left ventricle; LA – left atrium; pMV – prosthetic mitral valve, AOc – aortic conduit; RVOT – right ventricle outflow tract; PA – pulmonary artery; pAV – prosthetic aortic valve; ePs – excluded pseudoaneurysm).

At 3 months after discharge the patient was evaluated, being clinically well, presenting fatigue only for larger efforts, NYHA functional class I-II, no signs of congestion and without other hospital admissions.

Discussion

It is a case of aortic PVE with significant damage to the aortic annulus and MAIVF, leading to a moderate-to-severe para-valvular leak and very large aortic pseudoaneurysm. TEE had a few more resolution than TTE in the diagnosis of the pseudoaneurysm, but the CT had the highest anatomic resolution characterizing the dimensions and localization of the pseudo-aneurysm, as well as evaluating possible complications as coronary compression, enhancing the importance of the multi-modality imaging.

After the surgical correction, a rare microbiological agent (*Kytococcus schroeteri*) was identified in the excised aortic mechanical prosthetic valve, using molecular tests (PCR 16s rRNA sequencing). The role of *Kytococcus* species in the pathogenesis of human invasive diseases is under-reported, which may happen

because of the frequent misidentification of these organisms as unspecified micrococci or environmental contaminants (Mnif *et al.*, 2006). However, with increasing awareness for this potential pathogenic agent and improving molecular techniques for its identification, *Kytococcus* species are likely to emerge as increasingly important bacterial pathogens (Mnif *et al.*, 2006). This case demonstrates the usefulness of sequencing of the 16S rRNA genes for the identification of *K. schroeteri*. Despite that, gram-positive cocci that are arranged in tetrads and that are oxacillin resistant and arginine dihydrolase positive should be recognized as potential *Kytococcus* species (Mnif *et al.*, 2006; Le Brun *et al.*, 2005)

A 2019's review of *K. schroeteri* IE reported cases, showed that all of them were prosthetic valve endocarditis, with very different periods between the valve implantation and the diagnosis of the PVE (minimum 8 months, maximum 10 years). In 9 cases, 7 involved an aortic valve and 2 involved a mitral valve (Al Tamtami *et al.*, 2019). *K. schroeteri* has also been associated with other site infections (pneumonia, septic arthritis and osteomyelitis), but only in immunocompromised patients, in contrast with the cases of PVE (Chan *et al.*, 2012; Amaraneni *et al.*, 2015; Hodiament *et al.*, 2010; Ha *et al.*, 2015). Based on this we can admit the HIV infection as a possible risk factor for *K. schroeteri* PVE, despite this association not being well established at the time.

Surgery is the recommended treatment for these cases. Regarding etiology, the isolation of *K. schroeteri* whether in the blood cultures or in the prosthetic removed material is very important, because of its antibiotic resistance profile. Triple therapy with vancomycin, rifampicin and gentamicin is the recommended regimen. The therapy duration is not well established in the literature, but several authors recommend long periods of antibiotic therapy, as a 6-week regimen (Liu *et al.*, 2012; Mnif *et al.*, 2006; Becker *et al.*, 2003).

Conclusions

A rare case of IE complicated by a very large aortic pseudoaneurysm is described, requiring surgery, due to a very rare agent which was not isolated in blood cultures in a HIV-positive patient with very mild features of infection. It is the first reported case of a *Kytococcus schroeteri* prosthetic valve endocarditis complicated by aortic pseudoaneurysm. This case provides insights about an emerging microbiological pathogen, for which there should be a high microbiological suspicion index (in the presence of gram-positive cocci that are arranged in tetrads and that are oxacillin resistant and arginine di-hydrolase positive) and has the potential to cause severe infections, including life-threatening complicated endocarditis, mainly in prosthetic valves. However, this clinical case also reinforces the good clinical response to the surgical and antibiotic treatment.

Conflicts of Interest: The Authors have no conflicts of interest

Ethical Approval: Written informed consent was obtained from the patient for publication

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