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A Rare Case of Non-Prosthetic Aortic Valve Infectious Endocarditis Caused by Achromobacter xylosoxidans

Authors' Contribution: Study Design A Data Collection B Statistical Analysis C Data Interpretation D Manuscript Preparation E Literature Search F Funds Collection G

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Conflict of interest: None declared

Patient: Male, 19-year-old **Final Diagnosis: Endocarditis**

Symptoms: Fever • weigh loss **Medication:**

Clinical Procedure:

Specialty: Cardiology

Objective: Rare disease

Background: Achromobacter xylosoxidans is a ubiquitous environmental gram-negative bacterium, very resistant to antibiot-

ics. Endocarditis caused by these bacteria is extremely rare, with only 20 cases described in the literature to our knowledge. Mortality rates are high, and treatment usually involves a combination of antibiotics and surgery. Nosocomial infections predominate with a strong association between bacteremia and immunosuppression.

Case Report: A 19-year-old immunocompetent male presented with endocarditis He had interatrial and interventricular

communication corrected at age 11 months and aortic coarctation correction at age 10. Initial echocardiogram showed a possible interventricular patch infection, which was later ruled out. He was treated initially for endocarditis with a combination of antibiotics, but because he remained febrile after appropriate antibiotic treatment, surgery was performed. The patient had a favorable outcome after surgery and was asymptomatic on

follow-up.

Conclusions: Endocarditis caused by A. xylosoxidans is extremely rare. To date, only 20 cases of IT have been reported in the

literature, of which only two involved a native valve. Given the scarcity of cases reported, there is no consen-

sus on the best treatment.

MeSH Keywords: Achromobacter denitrificans • Endocarditis, Bacterial • Heart Valves

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Background

Achromobacter xylosoxidans is a ubiquitous, environmental gram-negative bacterium, very resistant to antibiotics. Endocarditis caused by these bacteria is extremely rare, with only 20 cases described in the literature to our knowledge. Mortality rates are high [1], and treatment usually involves a combination of antibiotics and surgery. Nosocomial infections predominate with a strong association between bacteremia and immunosuppression [2]. Here, we present a case of endocarditis caused by these uncommon bacteria in the native valve in an immunocompetent patient with no recent hospitalizations and a history of cardiac surgery, who survived after a combination of surgical and antibiotic treatment.

Case Report

A 19-year-old man presented with a history of approximately 1 month of intermittent fevers, chills, dry cough, and pleuritic chest pain. He lost about 10 lb during this period and had progressive fatigue. He had a medical history of interatrial and interventricular communication corrected at 11 months of age and aortic coarctation correction at age 10.

On examination, the patient was febrile, tachycardic, and his blood pressure was 125 over 76 mmHg. Physical exam demonstrated diastolic aortic murmur, grade 3/6, compatible with aortic insufficiency. There was no jugular vein distention, the lung exam was clear, and there were no petechiae or signs of skin lesions. The rest of the patient's physical exam was normal. Laboratory testing revealed an elevated white blood cell count of 16.45 109/L and microcytic anemia with hemoglobin of 9.1 g/dL and mean corpuscular volume of 78 fL. Creatinine was 0.71 mg/dL with a glomerular filtration rate above 60 mL/min. An HIV test done on our patient was negative. Chest x-ray showed signs of mild pulmonary congestion and an enlarged cardiac silhouette. An echocardiogram revealed a moderate left atrium enlargement and eccentric left ventricular hypertrophy with an ejection fraction of 47%. A filamentous structure was also seen inside the patient's left ventricle, adherent to a bicuspid aortic valve, and the ventricular patch in his inter-ventricular septum and severe aortic insufficiency. A diagnosis of endocarditis was given and the patient was placed on gentamicin, oxacillin, and ceftriaxone.

Two sets of blood cultures collected on the patient's admission were positive for presence of *Achromobacter xylosoxidans* sensitive only to carbapenem with minimum inhibitory concentrations (MIC) of 0.5 and resistant to oxacillin and ceftriaxone MIC 16 for both. The antibiotics were switched but the patient continued to have fevers after 7 days of appropriate antibiotics associated with increased shortness of breath and lung

congestion compatible with acute heart failure. Subsequent blood cultures performed on Day 5 of antibiotics were still positive. Because of the patient's clinical non-response and acute-onset heart failure associated with aortic insufficiency, a surgical procedure was scheduled [3].

During the surgery, vegetation was seen on the patient's aortic valve but not on his ventricular patch. Culture of the vegetation showed *A xylosoxidans*. The surgery was done with no major complications. He went to intensive care on moderate doses of vasopressors and was extubated 12 hours after the procedure.

On the fourth day of the patient's hospitalization, he had a spontaneous right-sided pneumothorax, with no apparent cause, which was drained and resolved after 3 days. He went home 10 days after surgery and completed 28 days of carbapenem. His immediate and 30-day echocardiograms at follow-up did not show any vegetation. He was symptomatic at his 6-month and 1-year follow-up appointments.

Discussion

Endocarditis is dangerous and difficult to treat. Prolonged fevers with weight loss in patients with a previous history of heart disease, especially after cardiac surgeries, should raise suspicion of this diagnosis.

A. xylosoxidans is an aerobic, motile, gram-negative rod that was first described in 1971 by Yabuuchi and Ohyama, who discovered it in patients with chronic, purulent otitis media [4]. Infections with A. xylosoxidans have included meningitis, pneumonia, peritonitis, and urinary tract infections [5,6]. However, bacteremia associated with prosthetic or native valve endocarditis caused by A. xylosoxidans is rare.

A recent review published by Barragan, et al. showed that in the majority of cases, patients were immunocompromised and almost all had acquired their infections in the hospital [7]. Reviewing our patient's records, he had no history of recent or recurrent illnesses or hospitalizations. In most patients, infections associated with this pathogen are related to catheters [7,8] but in our report, we do not have a clear source of his infection.

To date, only 20 cases (Table 1) of *A. xylosoxidans* endocarditis have been reported in the literature, of which only two involved a native valve [8–28]. Considering the treatment and outcome: 11 of 19 (58%) required surgical intervention; 8 of 17 (47%) died, 2 of the 8 deaths (22%) were from the operated group and the other 6 (75%) were from the clinically treated group [10].

Given the scarcity of the cases reported, there is no consensus on the best treatment for *A. xylosoxidans* endocarditis. Antipseudomonal penicillin and carbapenems are the best choices, based on bacteriologic studies and case reports [11].

In contrast to use of a percutaneous atrial septal occluder device, surgical patch closure of atrial septal defects is known

to represent no risk infective endocarditis [12]. To our knowledge, only two cases of endocarditis on a surgical patch of a ventricular septal defect have been reported [12]. Considering these previous case reports, it would be unlikely for our patient to have an infection in his patch. However, his bicuspid aortic valve increases his risk of developing endocarditis.

Table 1. Reported cases of IE.

Author	Age	Risk of IE	Comorbidities	Valve	Prosthetic	Antibiotic	Surgery	Died
This case	19 y	CS, Bicuspid aortic valve	None	Ao	No	Meropenem	Yes	No
Levoy et al. [8]	6 m	IVC+ calcified MV	Arterial calcification	M	No	Piperacillin-tazobactam+ TMP-SMX+colistin+ meropenem+levofloxacin	No	No
Tea et al. [9]	67 y	Rheumatic mitral stenosis, asplenia	Asplenia	M	No	Piperacillin-sulbactam+ Imipenem	Yes	NA
Rodrigues et al. [10]	86 y	None	IHD, lung fibrosis, CKD, plymyalgia	Ao	No	Piperacillin-tazobactam+ TMP-SMX	No	No
Derber et al. [11]	54 y	PV+ Fallot's T	Fallot's T	Р	Yes	Piperacillin-tazobactam+ Imipenem-Cilastatin Levofloxacin	Yes	No
Kumar et al. [13]	54 y	NA	CKD, CRF, H	M+Ao	No	Vancomycin+piperacillin- tazobactam+gentamicin	Yes	NA
Rafael et al. [14]	50 y	CS	VSDR	P+RVOT	No	NA	Yes	No
Sawant et al. [15]	62 y	PV+PM	AF, CHF, COPD, CKD	M+Ao+PM	Yes/No/–	Piperacillin-tazobactam+ TMP-SMX+amikacin+ meropenem+rifampicin	Yes	No
Tokuyasu et al. [16]	86 y	PV	NA	Ao	Yes	Carbapenem	No	Yes
Store et al. [17]	79 y	None	H, AF, TIA	M+Ao	No	Meropenem	No	Yes
Malek-Marín et al. [18]	50 y	Catheter	CKD	NA	_	NA	Yes	Yes
Ahmed et al. [19]	69 y	PV	DM, H, CKD, CABG	M+Ao	No/Yes	Ertapenem+Tigecycline+ TMP-SMX	Yes	Yes
van Hal et al. [20]	37 y	PV+IDU	NA	Ao	Yes	Carbapenem	Yes	No
Yang et al. [21]	35 y	IDU+TR+MP	Hepatitis C	T	No	Piperacillin-tazobactam+ Amikacin+Ceftazidime	Yes	NA
Nanuashvili et al. [22]	46 y	None	DM, Emphysema, IS	M+Ao	No	Ampicillin+Sulbactam+ Cotrimoxazole	Yes	No
Ahn et al. [23]	35 y	CS+PM	VSDR, CHB with PM	PM+RVOT	-	Ceftazidime+Amikacin	Yes	No
Martino et al. [24]	33 y	IVC	Bone marrow transplant	NA	-	Aztreonam+Amikacin	No	Yes
Davis et al. [25]	30 y	NA	HF	NA	_	None	No	Yes

Table 1 continued. Reported cases of IE.

Author	Age	Risk of IE	Comorbidities	Valve	Prosthetic	Antibiotic	Surgery	Died
Lofgren et al. [26]	77 y	PV	Rheumatic dis.+ PV	M+Ao	No/Yes	Tobramycin+Carbenicillin+ TMP-SMX+Moxalactam	- No	Yes
Bhattarai et al. [27]	37 y	IDU+PV	NA	М	Yes	Meropenem	Yes	No
Olson et al. [28]	35 y	Aortic surgery+PV	NA	Ao	Yes	Carbenicillin+ TMP-SMX+ Rifampicin+ Moxalactam+Azlocillin	No	Yes

AF – atrial fibrillation; Ao – aortic; CABG – coronary artery bypass grafting; CHB – complete heart block; CHF – congestive heart failure; CKD – chronic kidney disease; COPD – chronic obstructive pulmonary disease; CS – cardiac surgery; DM – diabetes mellitus; H – hypertension; HF – heart failure; IDU – intravenous drug user; IHD – ischemic heart disease; IS – ischemic stroke; IVC – intravenous catheter; M – mitral; NA – not available; P – pulmonary; PM – pacemaker; PV – prosthetic valve; RVOT – right ventricular outflow tract; T – tricuspid; TIA – transient ischemic accident; TMP-SMX – trimethoprim-sulfamethoxazole; TR – tricuspid regurgitation; VSDR – ventricular septal defect repair.

Conclusions

We presented an uncommon and rare case of *A. xylosoxidans* as the cause of infective endocarditis in a young, immunocompetent patient. The bacteria is very resistant and the treatment usually requires broad-spectrum antibiotics associated with surgical procedures. Our patient had a good outcome and was asymptomatic in later follow up.

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