

Case Report

Mycotic Pulmonary Artery Aneurysm Following Operation for Mitral and Tricuspid Valve Endocarditis in a Patient with Unrepaired Ventricular Septal Defect

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Abstract

A 26-year-old woman with unrepaired Ventricular Septal Defect (VSD) presented with dyspnea and fever and she experienced recurrent pneumonia for 6 months. The transthoracic echocardiography showed a perimembranous VSD, multiple mobile vegetation at mitral and tricuspid valve leaflet, dilated right atrium and ventricle and severe tricuspid regurgitation. Blood culture revealed the causative pathogen of *Streptococcus sanguinis*. A CT scan revealed multiple thromboembolism in pulmonary artery of right and left upper lobe. Operation of VSD closure and tricuspid and mitral valve replacement using mechanical valve was performed immediately. After that, subsequent serial imaging revealed the rapid enlargement of one of the aneurysms and possible rupture which proved to be fatal. We underwent digital subtraction arteriography to delineate Pulmonary Artery (PA) aneurysm *via* femoral vein approach. It revealed huge left interlobar PA aneurysm with wide opening. Because of mechanical tricuspid valve replacement, transcatheter embolization of mycotic PA aneurysm was not possible. Operation of left PA aneurysmectomy, left PA angioplasty and left lingular segmentectomy was performed *via* left thoracotomy. After surgical management, she discharged 4 weeks later without complication.

Keywords: Mycotic aneurysm; Ventricular septal defect; Infective endocarditis

Introduction

A mycotic aneurysm is an aneurysmal dilatation of arterial vessels caused by blood stream infection or septic emboli. Mycotic aneurysm involving the Pulmonary Artery (PA) is rare but fatal because degenerative vessel wall secondary to bacterial infection led to enlarged aneurysm prone to rupture [1-3]. Regarding the high mortality rate associated with aneurysmal rupture, treatment options tend to favor endovascular embolization and surgical approach such as lobectomy rather than conservative management [4-6].

Here, we report a case of multiple mycotic PA aneurysm secondary to infective endocarditis in a female patient with an unrepaired Ventricular Septal Defect (VSD), which was successfully treated with surgical intervention.

Case Presentation

A 26-year-old woman with a medical history of unrepaired VSD presented with dyspnea and fever for 2 weeks. She had history of recurrent pneumonia for 6 months at other hospital. On physical

examination, there were multiple edema and millet-sized petechia in both legs. Chest radiography performed in the emergency room showed increased opacity in left lower lung and right middle lung zone. The transthoracic echocardiography showed a perimembranous VSD, multiple mobile vegetation at mitral and tricuspid valve leaflet, dilated right atrium and ventricle and severe tricuspid regurgitation (Figure 1). The C Reactive Protein (CRP) was 15.80 mg/dL (normal 0.0~0.5 mg/dL), procalcitonin was 5.29 ng/mL (normal 0.0~0.05 ng/mL), Cardiac enzyme workup was also performed, hs-troponin 0.030 ng/mL (normal 0.000~0.014 ng/mL), NT-proBNP 10150 pg/mL (normal 0~115 pg/mL), and CK-MB was normal. The rheumatologic workup including anti-nuclear antibody, anti-neutrophil cytoplasmic antibodies, and anti-DNA antibody were all negative. The patient was admitted to the ICU and blood culture revealed the causative pathogen of *Streptococcus sanguinis* and started on third generation cephalosporin. A chest computed tomography scan revealed multiple thromboembolisms in pulmonary artery of right and left upper lobe (Figure 2). Operation of VSD closure and tricuspid and mitral valve replacement using mechanical valve was performed immediately. After that, subsequent serial imaging revealed the rapid enlargement of one of the aneurysms and possible rupture which proved to be fatal. We underwent Digital subtraction arteriography to delineate PA aneurysm *via* femoral vein approach. It revealed huge left interlobar PA aneurysm with wide opening (Figure 3). Because of mechanical tricuspid valve replacement, transcatheter embolization of mycotic PA aneurysm was not possible. Operation of left PA aneurysmectomy, left PA angioplasty with bovine pericardium and left lingular segmentectomy was performed *via* left thoracotomy (Figure 4). She discharged 4 weeks later after second operation. During follow-up for 1 year, transthoracic echocardiography showed mild mitral and tricuspid valve regurgitation, and chest radiography showed improvement of septic embolisms in both lungs, and she is free from symptom.

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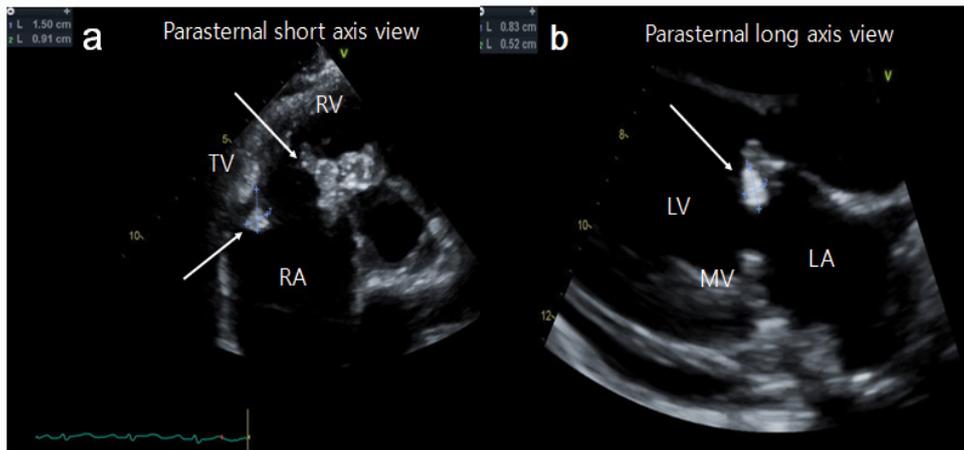


Figure 1: Transthoracic echocardiography showing multiple vegetation. Arrows indicate multiple vegetation on tricuspid valve annulus (a) and mitral valve annulus (b) MV: Mitral Valve; LA: Left Atrium; LV: Left Ventricle; RA: Right Atrium; RV: Right Ventricle; TV: Tricuspid Valve

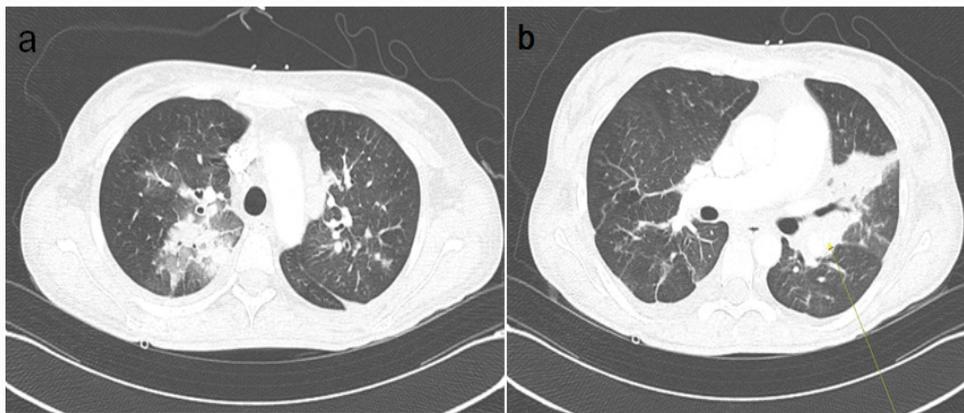


Figure 2: Lung CT scan showing multifocal thromboembolism in segmental pulmonary artery of right upper lobe (a) and left upper lobe (b). Arrow indicate poorly defined nodule like thromboembolism.

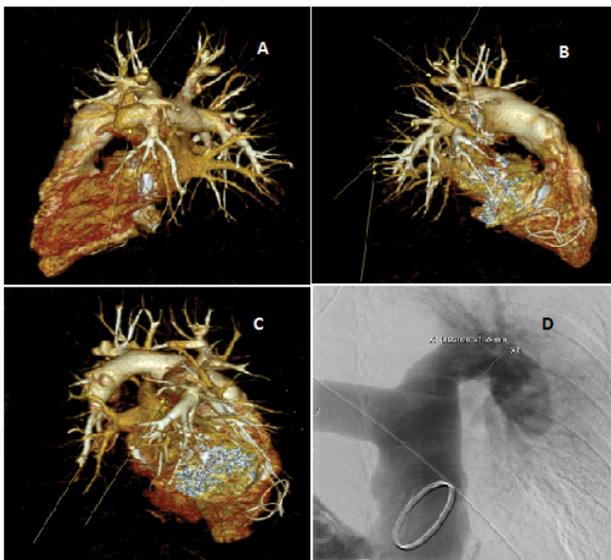


Figure 3: Cardiac CT and digital subtraction arteriography showing mycotic pulmonary artery aneurysm. CT reconstruction (A) shows multiple mycotic aneurysm of left and right pulmonary artery (arrows). Digital subtraction arteriography (B) shows huge mycotic aneurysm with 21 mm sized opening of left pulmonary artery (arrow).



Figure 4: Intraoperative finding of mycotic pulmonary artery aneurysm and resection specimens Left pulmonary artery was seen via left thoracotomy (A). Aneurysmectomy was performed between distal left pulmonary artery and proximal aneurysm. And then vascular sling was applied (B). Left pulmonary angioplasty was performed using bovine pericardium (C). The aneurysmal tissue specimen of pulmonary artery was seen (D and E).

Discussion

Mycotic PA aneurysm is a rare complication of right-sided endocarditis and correlated with Congenital Heart Disease (CHD). The proposed mechanism for infection dissemination from bacterial endocarditis is direct extension into the vessel wall from intraluminal septic thromboembolus, or hematogenous spread [1,7]. The common pathogen is known as *Staphylococci* and *streptococci*, but mycobacterial and fungi have been reported [6].

The management of mycotic PA aneurysm is difficult because there is no clear guideline about surgical intervention. As in our case of multiple and gradually enlarged aneurysms immediately after surgical correction of VSD and mechanical tricuspid valve replacement, the management is more difficult and challenging. Traditionally conservative management with antibiotic therapy and surgical therapy has been the main treatments. Recent report suggested that for clinically stable patients with mycotic PA aneurysm without sign of rupture and no trend of aneurysm dilatation, conservative therapy is considered instead of aggressive intervention [8]. Surgical management include aneurysmectomy, lobectomy, pneumonectomy or embolotherapy [4,6,7].

As an alternative to surgical treatment, aneurysm embolization can be performed using coil and vascular plug *via* endovascular approach. In our case, percutaneous embolization was impossible because we could not access to the PA due to previous mechanical tricuspid valve replacement.

Recent report demonstrated successful embolization using the balloon occlusion technique with the injection of ethylenevinyl alcohol copolymer in a pediatric patient with mycotic PA aneurysm [5]. Mycotic aneurysm has a possibility of rupture due to the fragility of vessels, so percutaneous embolization should be performed with caution [5].

Conclusion

In the study, we should be aware of the potential for mycotic PA aneurysm following infective endocarditis in patients with unrepaired CHD. In addition, the possibility of mycotic PA aneurysm should always be considered if there are findings of artery emboli in an imaging study with signs of infection. Although the mycotic PA aneurysm is very rare, rapid identification and adequate treatment is critical to prevent life-threatening complications in patient with unrepaired CHD.

Authors' Contributions

HS drafted the manuscript and carried out literature search and analysis. JB critically revised the manuscript and approved the final manuscript.

Ethics Approval Statement

This study was performed in accordance with the Helsinki Declaration and ethics approvals were obtained from the Institutional Review Boards of the authors' hospital.

Patient Consent Statement

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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