Case Report

Pulmonary endarterectomy for saddling pulmonary embolism by Aspergillus fungus in an immunocompetent patient

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A B S T R A C T

We present a case of Tricuspid valve Aspergillus endocarditis with saddle shaped massive pulmonary embolism occurring in an immunocompetent host. The patient was managed uniquely by pulmonary endarterectomy (PEA) and combination antifungal chemotherapy with Liposomal amphotericin-B + caspofungin.

1. Introduction

Fungal endocarditis is uncommon and accounts for less than 10% of all cases of infective endocarditis. Fungal endocarditis is related usually to intravenous drug abuse, cardiac prosthesis or an immunocompromised state. Aspergillus species causes approximately 20–30% of all fungal endocarditis cases while rests of the cases are contributed usually by Candida species.1 Here we present the case of complicated right sided Aspergillus endocarditis in an immune host that was encouragingly managed by synergistic antifungal drugs and PEA.

2. Case report

A 23-year-old female was referred for cardiac evaluation in view of moderate to high grade fever since past 1 month and worsening dyspnea from New York heart association (NYHA) class II to NYHA class IV over the past 15 days. The patient's temperature was 101 °F and blood pressure was 100/48 mm Hg. Her heart rate and respiratory rate were 110 beats/min and 28/min respectively. On physical examination the patient had pallor and a prominent V-Y descent waveform in Jugular venous pulse. No clubbing, or peripheral stigmata of endocarditis were present on physical examination. The resting
The patient was then shifted for emergency surgery. A restrictive perimembranous ventricular septal defect (VSD) (<1/3rd of the size of aortic annulus) with tricuspid valve impinging on it. Large thrombus adherent to the pulmonary valve, extending in to the main pulmonary artery (MPA) and its bifurcation causing severe pulmonary stenosis of 100 mm Hg gradient with streaky antegrade flow (Fig. 1B, C, D). The right ventricle and conus were free from the vegetation. The left sided structures were normal.

After obtaining multiple blood samples for cultures, the patient was started on empiric vancomycin and amikacin. Patient then underwent spiral chest computed tomography with contrast enhancement that showed filling defects in MPA with contrast enhancement that showed filling defects in MPA and its bifurcation by a saddle shaped thrombus (Fig 2A). An alveolar opacity was noted in the lower left lung parenchyma suggestive of consolidation with syn-pneumonic effusion. The patient continued to have fever in the postoperative period. Liposomal Amphotericin-B (3 mg/kg/day) was added by 50 mg daily thereafter as i.v. infusion) was added.

3. Surgical and postoperative course

1. Dilated right atrium, right ventricle, and large (2.5 x 2.0 cm) mobile vegetation attached to the septal leaflet of the tricuspid valve causing severe regurgitation (Fig 1A).

2. A restrictive perimembranous ventricular septal defect (VSD) (<1/3rd of the size of aortic annulus) with tricuspid valve impinging on it.

3. Large thrombus adherent to the pulmonary valve, extending in to the main pulmonary artery (MPA) and its bifurcation causing severe pulmonary stenosis of 100 mm Hg gradient with streaky antegrade flow (Fig. 1B, C, D). The right ventricle and conus were free from the vegetation. The left sided structures were normal.

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4. Discussion

The optimal treatment approach for fungal endocarditis requires aggressive surgical debridement in conjunction with antifungal therapy. Our patient presentation, echocardiographic findings and subsequent management were unique on the following grounds:

1. Congenital heart disease is a substrate for infective endocarditis in 10–20% of young adults. The absence of neutropenia; steroid abuse; intravenous drug abuse; lymphoid or leukemic disease led us to the presumption of normal immune status of the patient with VSD being the only predisposing defect. A recently published case report also describes A. fumigatus endocarditis in an immunocompetent patient. While this report does not recommend PEA due to inability to find a cleavage plane, we were able to dissect into the intimal layer. With endarterectomy we were able to remove the vegetation enbloc while an attempt at piecemeal removal would have resulted in embolisation or incomplete debridement. As the vegetation cast also extended into the bifurcation we could not resort to the technique of pulmonary root replacement that is described for isolated pulmonary valve endocarditis. Voriconazole is approved for first-line therapy in Invasive aspergillosis. Not only because of availability in our set up, we used the combination of liposomal amphotericin B and caspofungin with the rationale of achieving synergism by inhibition of fungal cell wall biosynthesis by caspofungin and loss of fungal cell membrane integrity by amphotericin B. A recent prospective study also showed promising results with such combination in invasive aspergillosis compared with monotherapy in immunocompromised patients.

2. PEA in infectious pulmonary embolism is rare. While a single report of PEA in infectious pulmonary embolism exists, the authors describe that intravenous drug abuse was the only predisposing factor. We believe that the patient had a large thrombus adherent to intima of MPA and branch pulmonary artery rather. There is a single report of PEA in infectious pulmonary embolism thus making our surgical management unique. While this report does not recommend PEA due to inability to find a cleavage plane, we were able to dissect into the intimal layer. With endarterectomy we were able to remove the vegetation enbloc while an attempt at piecemeal removal would have resulted in embolisation or incomplete debridement. As the vegetation cast also extended into the bifurcation we could not resort to the technique of pulmonary root replacement that is described for isolated pulmonary valve endocarditis. Voriconazole is approved for first-line therapy in Invasive aspergillosis. Not only because of availability in our set up, we used the combination of liposomal amphotericin B and caspofungin with the rationale of achieving synergism by inhibition of fungal cell wall biosynthesis by caspofungin and loss of fungal cell membrane integrity by amphotericin B. A recent prospective study also showed promising results with such combination in invasive aspergillosis compared with monotherapy in immunocompromised patients.
Fig. 1  – (A) Apical 4 chamber view showing large mobile vegetation on septal leaflet of tricuspid valve. RA = right atrium; RV = right ventricle. (B) Suprasternal view showing fungal mass almost completely filling the main pulmonary artery. (C) Parasternal short axis view showing saddle shaped fungal mass in pulmonary artery. RVOT = right ventricular outflow tract; Ao = aortic rim. (D) Severe pulmonary stenosis of ~ 100 mm Hg across the pulmonary valve.

Fig. 2  – (A) Pre operative 3D-CT angiogram showing filling defects in MPA and its bifurcation (arrow). (B) Following atriotomy, friable, bulky vegetation attached to the tricuspid valve (arrow). (C) RVOT incision extended into MPA showing embolus in MPA (arrow). (D) Y-shaped fungal cast removed after pulmonary endarterectomy.
5. Conclusion

An immunocompetent host, aggressive surgical debridement and combination antifungal therapy for invasive aspergillosis resulted in favorable prognosis in our patient. Large vegetation can embolise to the pulmonary vasculature creating severe pulmonary stenosis that requires emergency removal by embolectomy or endarterectomy.

Conflicts of interest

All authors have none to declare.

REFERENCES


