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CASE REPORT

Ruptured left ventricular subvalvar mitral aneurysm into the left atrium and left ventricle to left atrium fistula: case report of two pathological entities

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Summary
Subvalvar mitral aneurysm is a rare entity that mostly occurs in the African population. We present a case of a 23-year-old male patient who presented with shortness of breath and atypical chest pain. On preoperative transthoracic echocardiography, a subvalvar mitral aneurysm was noted with severe regurgitation from the aneurysm opening into the left atrium. The surgical findings revealed a single aneurysm neck present in the posterior mitral valve annulus. In addition to and separate from the aneurysm, a left ventricular to left atrial fistula was present.

Keywords: subvalvar mitral aneurysm, ruptured left ventricle, left atrium fistula

Introduction

Subvalvar mitral aneurysm is a rare entity and occurs most commonly in the African population. A predisposition or a congenital cause is usually responsible for the disease. Subvalvar mitral aneurysm was first described by Corvisart in 1812.[1] Subaortic aneurysms usually occur in relation to the left coronary cusp of the aortic valve. Submitral aneurysms are more common and usually occur in relation to the posterior annulus of the mitral valve.[2] These patients usually present with signs and symptoms associated with mitral valve regurgitation. In a case series of 12 patients reported by Du Toit et al., two patients had lesions suggestive of associated active rheumatic carditis and two patients had histological evidence indicative of tuberculosis.[2] An association with HIV, tuberculosis and rheumatic heart disease has been described.[3] Cases of rupture into the left atrial cavity have been resported.[4]

Ethics approval to report this case has been obtained from the Health Sciences Research Ethics Committee (HSREC) of the Faculty of Health Sciences, University of the Free State (reference number UFS-HSD2020/0027/3006). The patient provided written informed consent for the publication of his clinical details and images.

Case presentation

A 23-year-old male patient presented with shortness of breath and atypical chest pain. He had no history suggestive of rheumatic fever. On examination, the patient was stable and comfortable in a supine position. His blood pressure and pulse rate in the supine position were 105/63 mmHg and 86 beats per minute, respectively. He had a hyperdynamic precordium with an apex beat palpable in the left 6th intercostal space, in the mid-clavicular line.

On preoperative investigation, the 12-lead electrocardiogram (ECG) showed V2-V4 T wave inversion. His infection markers were elevated with a white cell count of 16 x 10^9/L and C-reactive protein (CRP) of 123 mg/L. On preoperative transthoracic echocardiography in parasternal long axis view, a giant (5.9 x 5.2 cm) submitral pseudoaneurysm was noted with severe regurgitation from the aneurysm opening into the left atrium (Figure 1). The mitral valve leaflets were visually normal with an enlarged left atrium (6.9 x 6.2 cm). A pericardial effusion of 2.6 mm to 8.4 mm with fibrin strands was noted. The patient had good global left ventricular systolic function with an ejection fraction of 66% calculated by means of modified Simpson's method.[5] He was presented for cardiac surgery to repair the aneurysmal defect.

The haemodynamic goals for the anaesthetic technique applied during the cardiac surgery were similar to a severe mitral regurgitation, to maintain forward flow and decrease the regurgitant fraction. Preload was maintained. We kept the heart rate high-normal at 80–100 beats per minute and avoided bradycardia. Sinus rhythm and contractility were maintained throughout the induction and maintenance periods. Afterload

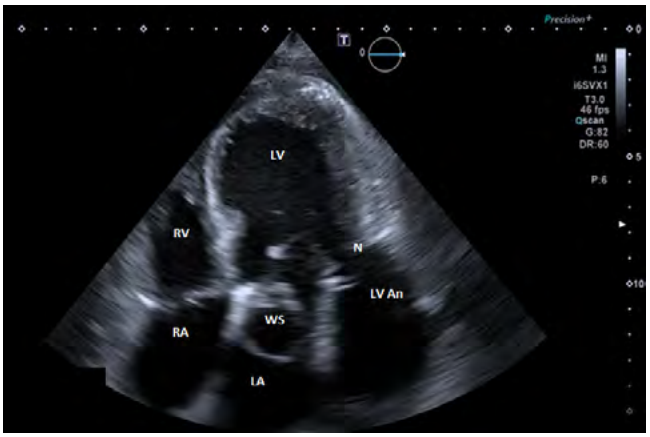

Figure 1: Transthoracic apical 4-chamber 2D view showing large aneurysmal sac with narrow neck and the windsock deformity opening into the left atrium. LV – left ventricle, N – neck of the aneurysm, LV An – left ventricle aneurysmal sac, WS – windsock deformity, RA – right atrium, RV – right ventricle

was reduced with sevoflurane to enhance forward flow. We avoided any increase in pulmonary vascular resistance.

Bispectral (BIS) and near-infrared spectroscopy (NIRS) in conjunction with routine South African Society of Anaesthesiology (SASA) monitoring[6] was used. Good peripheral venous access was secured with 16G and 14G cannulae and was applied pre-induction with the addition of an arterial line. Adrenaline was selected as the inotrope of choice and was started on induction at 0.03 µg/kg/minute. After a cardiostable induction and intubation, a central venous line was inserted into the right internal jugular vein using ultrasound guidance and transoesophageal echocardiography was performed. Anaesthesia was maintained with sevoflurane at a minimum alveolar concentration (MAC) of 0.8–1.2 and titrated to a BIS value of 40–60. The systolic blood pressure was kept in the preoperative range of 100–110 mmHg. Remifentanil was the opioid of choice and was infused with the Minto pharmacokinetic model using a target-controlled infusion (TCI) pump and titrated to effect, with an effect site concentration of 3–6 µg/ml.

A comprehensive transoesophageal echocardiogram examination was performed to show detailed images of the aneurysm to the surgeon in order to assist his surgical approach and technique. A giant submitral pseudoaneurysm with a neck of 8.3 mm that compressed the left atrium, with a windsock that opened up in the left atrium was clearly visible (Figure 2). Only one opening into the aneurysm and one exit from the aneurysm were visible. No clots were visualised. It could be observed in systole that the blood that entered the aneurysm, exited with high pressure into the left atrium. It was again confirmed that the mitral valve was unaffected and no associated mitral regurgitation was noted.

A diffusely thickened and inflamed pericardium with a fibrinous epicardial deposit was present, as shown in Figure 3. Intracardiac pathological findings were consistent with a large submitral left ventricular aneurysm with minimal thrombus load. A single aneurysm neck was present in the posterior mitral valve annulus

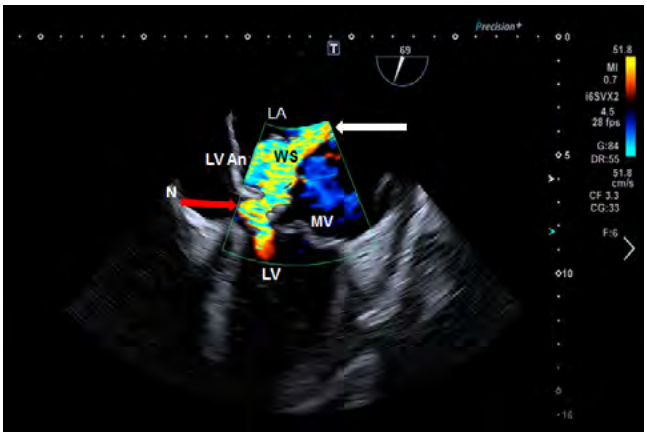

Figure 2: Mid-oesophageal mitral commissural view showing the aneurysm tunnelling through the submitral space into the left atrium with systolic regurgitation jet through the ruptured site (white arrow). The aneurysm with its narrow neck is clearly visible (red arrow). LA – left atrium, LV – left ventricle, LV An – left ventricle aneurysmal sac, WS – windsock deformity, MV – mitral valve, N – neck of the aneurysm

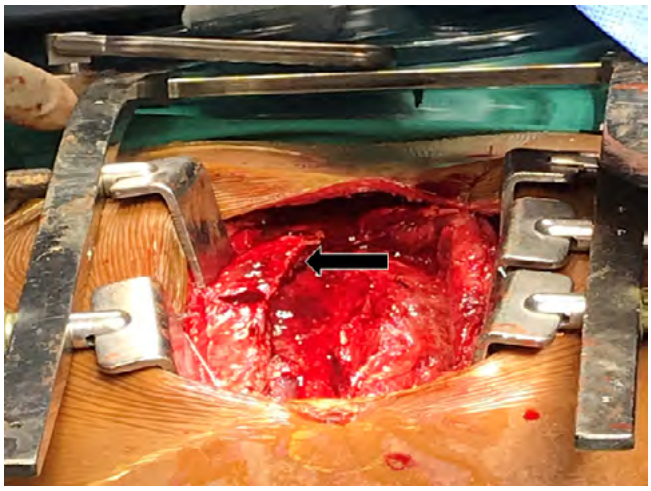

Figure 3: Thick and inflamed pericardium (arrow).

extending from the juncture of the P1/P2 scallops to the mid-P3 scallop level. The maximal depth of the neck of the aneurysm was 1 cm.

In addition to and separate from the aneurysm described above, a left ventricular to left atrial fistula was present. The 5 mm diameter opening was situated adjacent to the posterior mitral valve annulus on the lateral aspect of the P3 scallop. The fistula had a windsock deformity identical to the deformity present in congenital sinus of Valsalva aneurysms. The windsock deformity was 3 cm long with a ruptured tip. No obvious signs of infection were present. The mitral valve itself was otherwise anatomically normal.

Surgical repair was performed on cardiopulmonary bypass with bicaval atrial cannulation and moderate hypothermia. After aortic cross-clamping, myocardial protection was provided with intermittent antegrade cold blood cardioplegia. The technique used to repair the aneurysm was based on the intracardiac approach described by Antunes.[7] The mitral valve was exposed by a left atrial incision posterior to Sondergaard's groove. The neck of the aneurysm was initially identified by retracting the posterior mitral valve leaflet and examining the ventricular aspect of the posterior mitral valve annulus. A 5 cm incision was then made in the floor of the left atrium, 1 cm posterior to and parallel to the posterior mitral annulus, to allow access to the aneurysm neck. The neck of the aneurysm was closed directly with pledget-supported interrupted 2-0 TiCron (Covidien, Minneapolis, MN, USA) horizontal mattress sutures placed through the edge of the aneurysm neck on the left ventricular edge and then through the posterior mitral annulus. The incision in the floor of the left atrium was closed with a double layer running 3/0 Prolene (Ethicon, Sommerville, NJ, USA) suture technique. Subsequently, the left ventricle to left atrium fistula was repaired by resection of the windsock deformity and direct closure of the fistula opening with a running 4/0 Prolene suture technique. No further intervention on the mitral valve was deemed necessary.

The surgical procedure was performed by an experienced cardiothoracic surgeon, but was technically difficult, with a cardiopulmonary bypass time of 142 minutes. Pericardial and

fistula tissue sent for histological and microbiology investigation revealed no signs of tuberculosis and no bacterial organisms were cultured.

After separation from cardiopulmonary bypass, cardiac echocardiography examinations were performed in detail to exclude any residual flow into the aneurysmal sac. Apart from only a trivial mitral regurgitation, no flow was visible. The remainder of the procedure was uneventful and the patient was transferred from theatre in a stable condition to the cardiothoracic intensive care with no inotropic support required.

Discussion

We performed a preoperative transthoracic echocardiography and were of the opinion that we were dealing with a single opening from the aneurysm into the left atrium. The intraoperative transoesophageal echocardiogram (TOE) confirmed the preoperative transthoracic findings and no involvement of the mitral valve was observed.

The surgical findings provided more clarity on the precise pathology of the pseudoaneurysm and the left ventricle to left atrium fistula, with the windsock opening into the left atrium. Although our echocardiographic findings differed from the surgical findings and a preoperative diagnosis of the double pathology could have provided better assistance to the surgeon, the management of the patient was not adversely affected.

Modified Simpson's method for determining stroke volume and cardiac output can be inaccurate due to the underestimation of left ventricular end systolic volume (LVESV) with an overestimation of left ventricular stroke volume. Three dimensional chamber size quantification would be the preferred method of choice to estimate the LVESV. It is very important to confirm no residual flow in the aneurysm sac and fistula once the patient is off cardiopulmonary bypass. This case highlights the importance of identifying the possibility of double pathology prior to the surgical procedure.

Real time three dimensional TOE facilitates improved communication between physicians due to the accurate anatomical display of pathologies.[8] In conclusion, Multiplane echocardiography and live 3D zoom and 3D full volume TOE could have better delineated the pathology, being the aneurysm neck and windsock opening.

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Conflict of interest

The authors declare no conflict of interest.

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Ethical approval

Written informed consent for publication of the patient's clinical details and images was obtained from the patient. The patient signed our Hospital Consent form for publication (HSREC Patient Consent for publication Template form 016).

Ethics approval to report this case has been obtained from the Health Sciences Research Ethics Committee (HSREC) of the Faculty of Health Sciences, University of the Free State (reference number UFS-HSD2020/0027/3006).

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