CASE REPORT

Fortune or misfortune: asymptomatic, delayed presentation of complete dehiscence of mechanical aortic valve conduit and pseudoaneurysm

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SUMMARY

Complete dehiscence of a composite aortic valve graft with pseudoaneurysm formation is a rare complication following aortic root replacement. This complication often takes place in the setting of acute graft infection and accompanies symptoms of heart failure, valve insufficiency or sepsis. We present a delayed, asymptomatic presentation of this complication in a young man with distant history of aortic root replacement and medically treated prosthetic valve endocarditis a year postoperatively. He had been non-adherent to warfarin over 10 years, but otherwise maintained a healthy life. After being lost to follow-up, he re-presented 12 years after the initial operation with new-onset seizures. Echocardiogram revealed complete dehiscence of a composite valved conduit at the proximal anastomosis site with a resultant large pseudoaneurysm. The patient underwent an urgent re-operation with resection of the pseudoaneurysm and insertion of a tissue valved conduit. He had an uncomplicated postoperative recovery and promised close follow-up on discharge.

BACKGROUND

Dehiscence of an aortic valved conduit and resultant pseudoaneurysm formation are extremely rare, but serious complications of aortic root replacement. Sites of pseudoaneurysm formation include the distal suture line, the coronary artery buttons and the proximal valve suture line.¹ The most common cause of pseudoaneurysm formation is infective endocarditis, but technical issues and recurrent aneurysmal dilation can also be etiologic factors.² If left unnoticed, there is a high risk of spontaneous rupture leading to immediate death and/or urgent surgery with high mortality.³ The following is a case of an otherwise healthy, active young man with complete proximal dehiscence of a mechanical aortic root replacement who presented with an unrelated symptom 12 years after being lost to follow-up from his initial operation.

CASE PRESENTATION

A 37-year-old man was brought to the emergency department after a witnessed generalised seizure. His medical history was significant for repair of an ostium secundum atrial septal defect during the first year of life. He subsequently underwent an aortic root replacement with a mechanical valved conduit at age 24 for an aortic root aneurysm. He reported being readmitted 1 year after his initial

surgery for endocarditis, which was treated with 2 months of intravenous antibiotics without another surgical intervention. On this presentation, he was afebrile with normal haemodynamic parameters. Examination was pertinent for a postictal mental status without focal neurological deficit, as well as a grade 4/6 systolic murmur along the left upper sternal border. Further history obtained after the recovery from his postictal state revealed loss of medical follow-up and non-adherence to anticoagulation for almost 12 years since his recovery from prosthetic valve endocarditis. He otherwise endorsed an active lifestyle as a construction worker and denied any dyspnoea on exertion, chest pain or syncope.

INVESTIGATIONS

Transthoracic echocardiography (TTE) was performed as a part of the initial work-up to rule out cardioembolic stroke in the setting of a new-onset seizure in a patient with a mechanical valve. The TTE showed an echogenic structure located in the proximal ascending aorta distal to a dilated aortic root without significant aortic insufficiency, suggesting dehiscence of the prosthesis (figure 1). No vegetations or thrombus were identified on the valve or within the heart. The left ventricular size and function appeared normal. A closer examination with transesophageal echocardiography (TEE) further revealed excessive excursion of the aortic root tissue that appeared nodular and irregular, concerning for infection of the prosthesis versus pseudoaneurysm (figures 2 and 3). The mechanical valve was located ~2 cm distal to native aortic annulus but appeared to be well seated at the proximal end of the conduit (figure 4). Although dilated, the right and left coronary buttons appeared intact. Pulsed-wave Doppler interrogation showed holodiastolic flow reversal in the descending aorta, without haemodynamically significant aortic insufficiency (figure 5). There was no dissection throughout the visible thoracic aorta, which had a grade II intimal thickening with atheroma. The preoperative CT scan paralleled the anatomic findings on TEE (figure 6). The patient was afebrile, and multiple blood cultures were without growth of any organisms. Serial CT scans of head failed to reveal infarct or haemorrhage.

TREATMENT

The patient was urgently taken to the operating room for reoperative aortic root replacement. The

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Figure 1 Parasternal long-axis view on the transthoracic echocardiogram suggests dehiscence of aortic valve prosthesis (arrow), distal to native aortic annulus (asterisk).

decision was made to use a biological valve given the patient's reluctance with anticoagulation and his unstable insurance status. Initial challenges in the operating room included extensive adhesions and scar tissues that were expected during redo sternotomy, as well as the position of right coronary artery abutting the sternum. Cardiopulmonary bypass was established through the right femoral vessels, and after deep hypothermic circulatory arrest, the pseudoaneurysm was entered and the aortic graft was clamped. Cardioplegia was then given at 60 min interval, and an left ventricle vent was placed through the right superior pulmonary vein. The distal anastomosis was intact, but the entire proximal suture line was dehiscd with all of the valve sutures torn out of the aortic annulus (figure 7A). There was a large pseudoaneurysm, which was containing the proximal dehiscence (figure 7B). The coronary buttons were intact. The proximal valve and aortic graft were resected after excising the coronary buttons. The aortic root was then replaced with a tissue aortic valved conduit composed of a bovine pericardial valve sewn inside a Gelweave Valsalva graft.

OUTCOME AND FOLLOW-UP

The patient's immediate postoperative course was complicated by another episode of generalised seizure of unknown origin. He was transferred to the intensive care unit on low-dose inotropic infusion and mechanical ventilator support. He was

Figure 3 High-esophageal view of the ascending aorta on the TEE, showing the mechanical aortic valve at the proximal end of the conduit (arrow) within pseudoaneurysm (double arrow). TEE, transesophageal echocardiography.

weaned off inotropic infusion and was successfully extubated on postoperative day 1. He otherwise had uneventful recovery and was discharged on postoperative day 8 with appropriate follow-up appointments. He stated that this was an eye-opening experience for him and promised that he would follow-up very closely as an outpatient.

DISCUSSION

Complete dehiscence of an aortic valve conduit with pseudoaneurysm formation is a rare complication that necessitates an urgent re-operation, given the high risk of spontaneous rupture.³ The time interval between the initial surgical procedure and discovery of this complication is variable but has been reported up to 17 years after the initial operation.⁴ There have been isolated reports of graft dehiscence and pseudoaneurysm formation in non-infectious⁵ and infectious settings,⁶ where the patients presented with symptoms of heart failure or sepsis. Endocarditis in patients with ascending aorta prosthetic graft is usually a late complication, with median time from initial surgery up to 24 months, and carries high mortality due to life-threatening complications such as heart failure, uncontrolled sepsis and aortic rupture. Current recommendation is surgical management, owing to high rates of reinfection when prosthetic

Figure 2 Biplane images of the pseudoaneurysm with irregular and nodular lumen (double arrow), housing dehiscd aortic valve prosthesis (single arrow).

Figure 4 Apical 5-chamber view on TTE showing complete dehiscence of aortic prosthesis and pseudoaneurysm (asterisk) ~2 cm distal to native annulus (arrow). TTE, transthoracic echocardiography.

Figure 5 (A) Pulsed-wave Doppler of the descending thoracic aorta showing holodiastolic flow reversal. (B) Midesophageal aortic valve long-axis view shows no significant aortic prosthetic valve regurgitation during diastole.

Figure 6 CT scan without contrast showing the mechanical aortic valve in a large pseudoaneurysm.

endocarditis is managed with antibiotics alone.⁷ Our case is notable for an asymptomatic, delayed presentation of pseudoaneurysm formation, more than a decade after the infection of the prostheses that was managed medically. Owing to the significant mortality and morbidity of urgent pseudoaneurysm repair,⁴ current guidelines recommend long-term, annual monitoring with CT scan after the initial surgical repair of an aortic root and/or ascending aortic aneurysm.⁸ Our case not only reiterates the recommendations of current guidelines, but also emphasises the importance of maintaining low index of suspicion in patients with previous aortic root replacement even for an atypical presentation. It also demonstrates the role of echocardiography as an excellent initial imaging modality to detect disarticulation of the prosthesis and pseudoaneurysm. Our patient was likely able to maintain an active lifestyle without symptoms, owing to the absence of ventricular dysfunction or significant prosthetic valve regurgitation. Of note, the patient's Doppler echocardiography demonstrated holodiastolic flow reversal in descending aorta, which is a surrogate marker for severe aortic insufficiency. Another known factor that prolongs diastolic flow reversal is reduced compliance and increased stiffness of the aorta.⁹ Holodiastolic flow reversal in our patient

Figure 7 (A) Dissected ascending aortic graft with a well-seated mechanical aortic valve (arrow). (B) Pseudoaneurysm (arrow) and right coronary artery annulus (asterisk) after the aortic graft and valve were removed.

can be explained by two potential mechanisms: (1) increased aortic stiffness after initial surgery suggested by intimal thickening of thoracic aorta on TEE and (2) enhanced reversal flow Doppler signal in the setting of exaggerated downward motion of the entire aortic arch as pseudoaneurysm flattens during diastole.

Contributors KTO was the main author of this case report. JD, the primary surgeon of the case, provided the intraoperative details, surgical images and revision of the manuscript. CT provided echocardiogram images, edited the manuscript and, most importantly, supervised the whole process of this case report.

Competing interests None declared.

Patient consent Obtained.

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Learning points

- ▶ Dehiscence of an aortic root graft and subsequent pseudoaneurysm formation are extremely rare but fatal postoperative complications of aortic root replacement.
- ▶ Based on the current guidelines, a previously repaired aortic root needs close, indefinite surveillance imaging with CT scan or MRI, especially following postoperative infection.
- ▶ Patients with dehiscence of an aortic root graft and pseudoaneurysm formation may remain asymptomatic in the absence of significant prosthetic valve regurgitation.
- ▶ Transthoracic echocardiography and transesophageal echocardiography can serve as an excellent initial imaging modality to assess the anatomy and function of the valved conduit.
- ▶ Holodiastolic reversal of flow in descending aorta may not be a specific parameter of severe aortic insufficiency in a patient with an aortic root graft, especially with a large pseudoaneurysm.

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