A Case of Left Ventricular Outflow Tract Pseudoaneurysm: An Incidental Finding Revealing Occult Infective Endocarditis

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INTRODUCTION

Left ventricular outflow tract (LVOT) pseudoaneurysm is an uncommon complication of aortic valve replacement and aortic root surgery. It most commonly occurs in the context of infection, although it can have a nonspecific, insidious clinical presentation, making detection difficult. We describe the case of a patient diagnosed with a large LVOT pseudoaneurysm. This case demonstrates potential diagnostic pitfalls in the detection of LVOT pseudoaneurysm and the benefits of multimodality imaging with transthoracic echocardiography (TTE), transesophageal echocardiography and computed tomographic aortography (CTA).

CASE PRESENTATION

A 68-year-old woman presented for TTE as part of annual surveillance after previous redo mechanical aortic valve replacement and aortic root replacement. Follow-up was routine, but the patient reported several weeks of increasing fatigue. Her initial operation was at 21 years of age for a prematurely stenosed bicuspid aortic valve, for which she received a "ball-in-cage" Starr-Edwards mechanical valve prosthesis. This required revision at 60 years of age for restenosis of her valve and intravascular hemolysis. She underwent a redo mechanical aortic valve replacement requiring 2-hour division of adhesions to establish aortofemoral bypass. This operation was complicated by a ventricular septal defect and deterioration of the native aortic root requiring ventricular septal defect repair and aortic root replacement. The aortic root replacement required telescoping down to the LVOT. The patient was pacemaker dependent following the second aortic valve replacement. Her medical history was notable for polymyalgia rheumatica, for which she took long-term low-dose (1 mg/d) oral prednisolone, as well as hypertension.

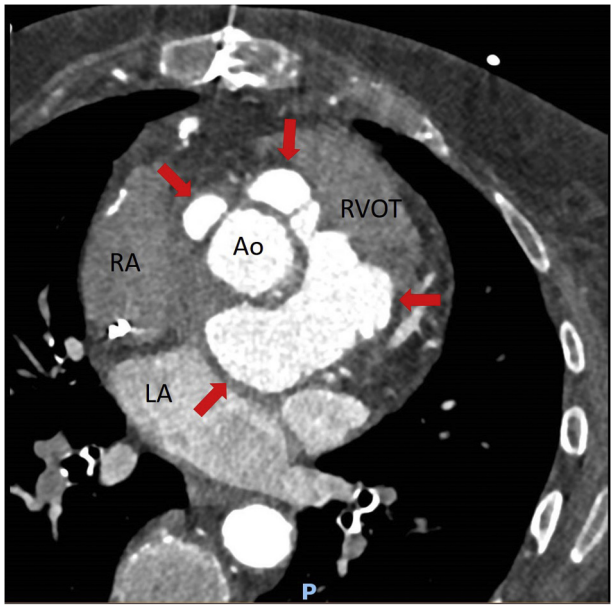

Figure 1 An axial slice from CTA above the level of the prosthetic aortic valve showing the proximal ascending aorta surrounded by the pseudoaneurysm cavity (*red arrows*). *Ao*, Aorta, *LA*, left atrium; *RA*, right atrium; *RVOT*, right ventricular outflow tract.

The patient's initial TTE revealed turbulent color Doppler flow around the prosthetic aortic valve (Video 1). This initially raised clinical suspicion for a severe perivalvular leak, with or without a possible LVOT pseudoaneurysm. The patient was sent for urgent CTA. This demonstrated a large aortic root pseudoaneurysm arising below the level of the prosthetic aortic valve (Figures 1-3), measuring 6.1 cm in anteroposterior diameter and 5.3 cm transversely with a craniocaudal extent of approximately 4.1 cm. The mechanical replacement aortic valve was shown to have dehisced from the outflow tract. However, in contrast to TTE, no paravalvular communication was found between the proximal ascending aorta graft and the subvalvular space, as the aortic valve and aortic root replacements remained contiguous. There was no free blood within the pericardial, mediastinal, or pleural space, likely because of dense adhesions.

The patient was admitted to hospital for urgent transesophageal echocardiography. The findings were notable for the aortic valve and root seen rocking to and fro within a large subvalvular pseudoaneurysm extending around the proximal aortic graft (Figure 4, Video 2). Color Doppler was used to define the limits of the aneurysm (Figure 5). The prosthetic aortic valve was well seated within the aortic

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VIDEO HIGHLIGHTS

Video 1: Original transthoracic imaging of the prosthetic aortic valve from the parasternal short-axis window. A hypolucent space is seen anterior to the prosthetic aortic valve. Color Doppler flow is evident around the valve, a chief differential diagnosis for which was aortic regurgitation due to valve dehiscence. Color Doppler flow is demonstrated in systole and diastole, more consistent with a pseudoaneurysm.

Video 2: Transesophageal echocardiography in the midesophageal three-chamber view with the use of x-plane to show the extent of the pseudoaneurysm. It is shown to extend medially, posteriorly, and laterally and to extend superiorly so that it is both subvalvular and supravalvular, surrounding the aortic root.

Video 3: Transesophageal echocardiography in the midesophageal three-chamber view with the use of color compare shows both a trivial perivalvular leak between the valve and aortic root prostheses and color Doppler flow within the pseudoaneurysm, which is contained.

Video 4: Transesophageal echocardiography in the transgastric position demonstrating trivial perivalvular leak between the valve and aortic root prostheses and color Doppler flow within the pseudoaneurysm.

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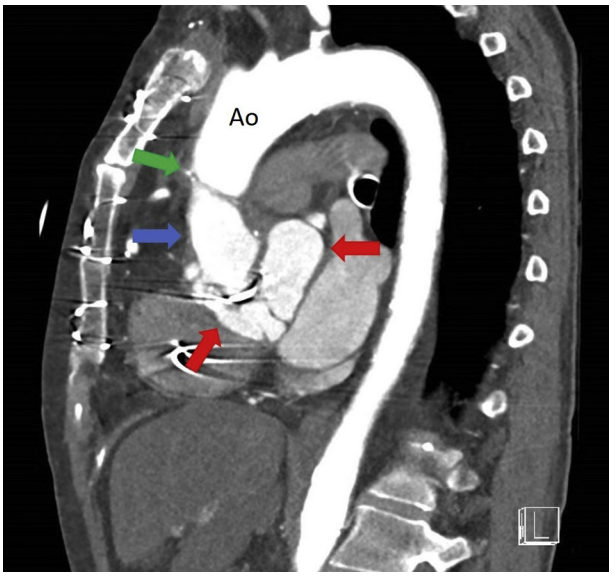

Figure 2 A sagittal slice from CTA showing the anterior and posterior extent of the pseudoaneurysm (*red arrows*) around the proximal ascending aorta prosthesis (*blue arrow*). The anastomosis between the prosthetic aortic root and the native aorta is visible (*green arrow*). *Ao*, Aorta.

graft, and there was only a trivial perivalvular leak on color Doppler (Videos 3 and 4). There was no evidence of vegetations associated with the prosthetic valve, the aortic graft, or any other native valves.

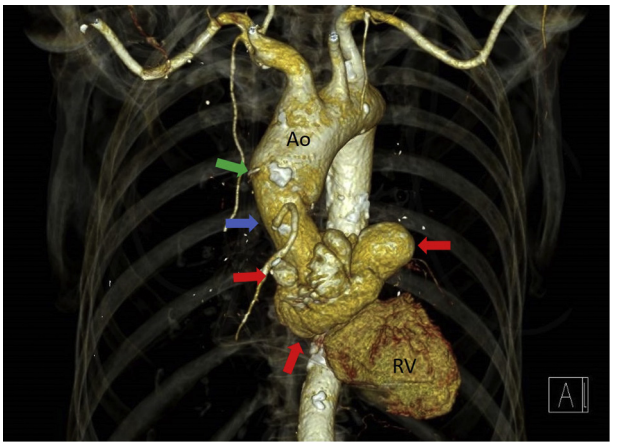

Figure 3 A three-dimensional reconstruction from CTA, viewed anteriorly, showing the extent of the pseudoaneurysm (*red arrows*), the prosthetic aortic root and proximal ascending aorta (*blue arrows*), and the anastomosis with the native aorta (*green arrow*). *Ao*, Aorta; *RV*, right ventricle.

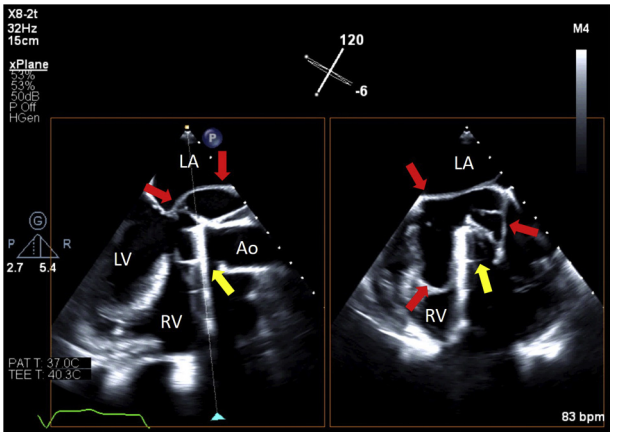

Figure 4 Transesophageal imaging from the midesophageal position, using x-plane to show the pseudoaneurysm in the long-axis (*left*) and short-axis (*right*) views. The extent of the pseudoaneurysm is demonstrated (*red arrows*) surrounding the prosthetic aortic valve (*yellow arrows*). *Ao*, Aorta; *LA*, left atrium; *LV*, Left ventricle; *RV*, right ventricle.

The patient underwent urgent redo sternotomy for aortic root replacement, aortic valve replacement, and repair of the pseudoaneurysm. The procedure was uncomplicated and the patient made a full recovery. Postoperative TTE showed a well-seated aortic valve prosthesis within the new aortic root graft and containment of the subvalvular pseudoaneurysm without flow evident on color Doppler (Figure 6).

Cultures of the aneurysm, LVOT, and old prosthetic valve were positive for *Enterococcus faecalis*. Tissue from the LVOT also grew *Aspergillus flavus*, felt to be nonpathogenic. Positron emission tomography showed no involvement of the permanent pacemaker or evidence of other systemic sites of infection. The patient was discharged from hospital on a 6-week course of benzylpenicillin and ceftriaxone. Clinical status 3 months after discharge was excellent.

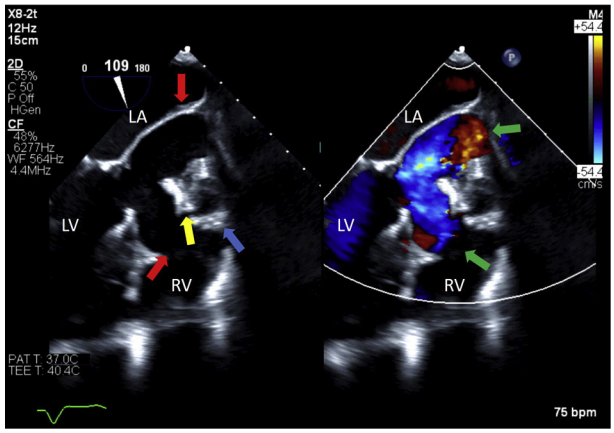

Figure 5 Transesophageal imaging from the midesophageal position. The *right panel* shows two-dimensional imaging in the long axis of the LVOT aneurysm (*red arrows*), prosthetic aortic valve (*yellow arrow*) and prosthetic aortic root (*blue arrow*). The *left panel* shows color compare, defining the extent of color Doppler flow (*green arrows*). *LA*, Left atrium; *LV*, Left ventricle; *RV*, right ventricle.

pathic LVOT pseudoaneurysm[6] and pseudoaneurysm caused by trauma[7] have also been reported.

Pathogenesis depends on the underlying cause. Where infection is implicated, it can be through hematogenous seeding of suture lines or by direct extension of infection from preexisting infective endocarditis.[8] Surgical suture techniques may also contribute, with tight sutures disrupting the LVOT wall and contributing to dehiscence. High flows in the LVOT cause further stress, as blood enters the surrounding mediastinum, forming a contained paravalvular leak.[9] One case-control series in patients without endocarditis showed that the implantation of composite grafts increases the risk for pseudoaneurysm compared with aortic valve replacement alone, though all pseudoaneurysms developed in this series were in the aortic root and ascending aorta, rather than the LVOT.[10] Our patient did not have prior infective endocarditis but was immunocompromised. Surgical cultures from the pseudoaneurysm were positive for *E faecalis*, suggesting that infection likely contributed to its formation.

The presentation of cardiac pseudoaneurysm can be varied, typically presenting with vague and nonspecific symptoms. Patients may present with chest pain or symptoms of heart failure, possibly due to local compression effect, impaired cardiac output, or left ventricular volume overload.[11] Our patient did not present with any defining

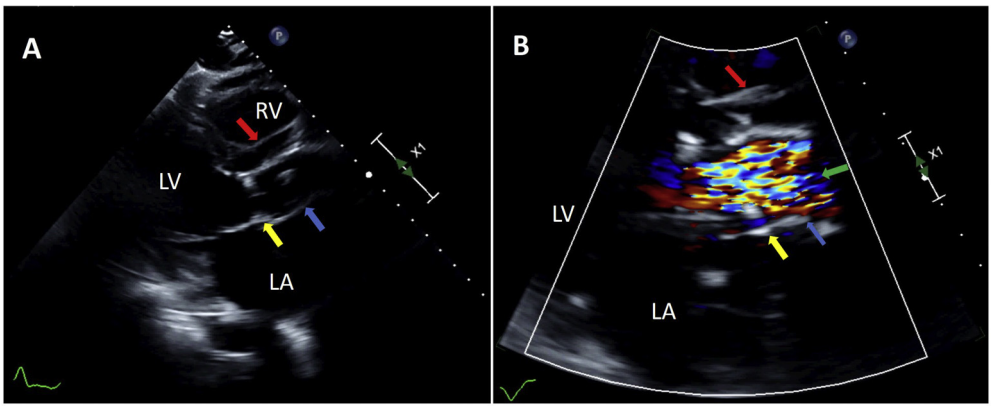

Figure 6 Transthoracic imaging was performed postoperatively. **(A)** A well-seated prosthetic aortic valve (*yellow arrow*) within the new prosthetic aortic root (*blue arrow*). The potential space created by the sealed pseudoaneurysm is visible (*red arrow*). **(B)** Color Doppler flow (*green arrow*) within the prosthetic aortic valve and root (*yellow and blue arrows*) but not within the old pseudoaneurysm (*red arrow*). *LA*, Left atrium; *LV*, Left ventricle; *RV*, right ventricle.

DISCUSSION

This case details the infrequent complication of LVOT pseudoaneurysm following mechanical aortic valve and aortic root replacement. After aortic valve surgery, 20% of late mortality is attributable to valve-related complications, with pseudoaneurysm being a known but uncommon complication.[1]

Pseudoaneurysm, or false aneurysm, is the rupture of an arterial wall, with blood contained by the outer adventitia layer. Cardiac pseudoaneurysm is most often seen in the left ventricle and is most commonly caused by myocardial infarction.[2,3] It is an important complication to recognize, with potentially fatal sequelae including rupture, infection, and thromboembolism.[4] A minority of cases are found in the LVOT. These are most often thought to be directly related to previous cardiac surgery or infective endocarditis.[2,5] Cases of idiosymptoms and despite operative evidence of infection did not display any other clinical features of endocarditis. The subacute nature of this patient's presentation is important and reinforces the need to thoroughly investigate for infective causes whenever LVOT pseudoaneurysm is detected.

Given its availability, low cost, and lack of radiation exposure, TTE is usually the first modality of choice to investigate cardiac pseudoaneurysm. Furthermore, given the often occult presentation, it can be argued that regular surveillance with TTE is a key strategy in the detection of both LVOT and other cardiac pseudoaneurysms. Kupari *et al.*[12] found that echocardiography, with cross-sectional and Doppler ultrasound studies, could identify left ventricular pseudoaneurysm following mitral valve replacement. However, the echocardiographic diagnosis is not without limitations, as evidenced in this case with

aortic valve dehiscence initially believed to be a possible differential diagnosis.

The use of other noninvasive modalities allows confirmation of the diagnosis and further characterization of the defect. These techniques include CTA and cardiac magnetic resonance imaging, with CTA anatomically defining the structural abnormality in this patient. Progressing to transesophageal echocardiography permitted further characterization of a large LVOT pseudoaneurysm, with Doppler demonstrating dynamic flow within the cavity. An accurate diagnosis is likely best achieved through an integrative multimodality approach.

There is a limited body of published evidence regarding the natural history of untreated LVOT pseudoaneurysm. The data that do exist come predominantly from single-center case series looking at all cardiac pseudoaneurysms. Frances *et al.*[3] found that medically treated left ventricular pseudoaneurysm had a 30% to 45% risk for rupture and a mortality rate of nearly 50%. Enseleit *et al.*[10] had a case-control cohort of noninfective pseudoaneurysms after aortic valve replacement and found that no patient needed reoperation over a follow-up period of several years. However, none of these patients had LVOT pseudoaneurysm, with all affecting the aortic root or proximal ascending aorta.

Given the possible complications, it is suggested that acute, enlarging, or symptomatic pseudoaneurysms require surgical repair.[13,14] With the now widespread availability of TTE, there is likely to be an increase in the detection of "incidental" pseudoaneurysms in asymptomatic patients. Our patient had minimal symptoms from the aneurysm, but given the size of the defect, she was planned for a third aortic valve replacement and surgical repair. Percutaneous closure of LVOT pseudoaneurysm has been described.[15] However, this is likely to be possible only in select cases with favorable anatomic characteristics and prohibitive surgical risk. In this case, with an extensive pseudoaneurysm that enveloped the previous prosthetic aortic root, percutaneous intervention was not feasible.

CONCLUSION

Cardiac pseudoaneurysm is an infrequent and potentially life-threatening condition. Pseudoaneurysm of the LVOT can occur after previous aortic valve or aortic root surgery and is often associated with infection. It can have an insidious clinical course, best detected through the use of routine surveillance TTE and, when discovered, is best characterized using a multimodality approach. Although percutaneous occlusion may be possible in a select number of cases, surgical intervention remains the preferred treatment.

SUPPLEMENTARY DATA

Supplementary data related to this article can be found at https://doi.org/10.1016/j.case.2020.05.008.

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