

Complex cardiac surgery in a high-risk patient with new-onset severe mitral regurgitation and aorta to right ventricular fistula after transcatheter aortic valve implantation: a case report

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Background

Transcatheter aortic valve implantation (TAVI) is the procedure of choice for aortic stenosis in high surgical risk patients, but it is no free from complications.

Case summary

A 86-year-old patient with severe aortic stenosis underwent TAVI 3 years ago with an Edwards Sapiens valve by femoral access. In the echocardiography follow-up, an aorta–right ventricular (Ao–RV) fistula was noted with restrictive flow and no significant shunt and it was treated conservatively. Three years after TAVI, the patient underwent cardiac surgery because of worsening heart failure due to a severe degenerative mitral regurgitation with tethering of P2 due to left ventricular remodelling, a posterior jet of severe regurgitation, and left ventricular dilatation. Surgical replacement of the TAVI and aortic root with a bioprosthesis (Medtronic Freestyle) and direct closure of the fistula was performed along with the mitral valve replacement. The patient was discharged with a good clinical result and no evidence of remaining Ao–RV fistula at transthoracic echocardiography.

Discussion

Aorta–right ventricular fistula is a rare entity. Most reported cases arise after rupture of a congenital coronary sinus aneurism, endocarditis, trauma, and aortic valve or aortic root surgery. This is the 10th reported case after TAVI (9 after an Edwards Sapiens TAVI). Non-significant shunt can be treated conservatively but development of heart failure and death are described in significant shunts. Balloon post-dilatation and the absence of surgical calcium debridement inherent to TAVI may theoretically contribute to the development of the fistula. Surgical replacement and closure of the fistula is a therapeutic option for this entity even in high-risk patients.

Keywords

Transcatheter aortic valve implantation (TAVI) • Aortocardiac fistula • Aortic valve replacement • Aorto–right ventricular fistula • Case report

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Learning points

- Complex patients should not be excluded from cardiac surgery only based on risk if a significant clinical benefit is expected and no less invasive comparable options are available.
- Aorto–right ventricular fistula is a rare complication of transcatheter aortic valve implantation.
- Non-significant shunt may be treated conservatively but important morbidity and mortality have been described in severe shunts.

Introduction

Transcatheter aortic valve implantation (TAVI) is the procedure of choice for aortic stenosis patients with high surgical risk and can be considered in intermediate and low-risk patients with favourable anatomy. While the procedure is less invasive than standard surgical valve replacement, it is not free from complications. In the follow-up of patients with aortic valve stenosis who were considered poor surgical candidates, incidental disease may pose again the indication for cardiac surgery.

The absence of debridement of the diseased calcified native aortic valve, which is inherent of the TAVI technique, has been related to paravalvular leaks, atrioventricular block, and vascular complications (including aortic root haematoma and rupture).^{1–3}

We expose the case of a patient who developed an aorta–right ventricular (Ao–RV) fistula shortly after transfemoral TAVI, initially conservatively followed, which developed degenerative mitral valve regurgitation and required subsequent cardiac surgery for mitral and coronary artery disease at the age of 86 years.

Timeline

3 years prior to surgery	Severe aortic stenosis TAVI (Edwards Sapiens valve) Aorto–right ventricular (Ao–RV) fistula noted on echocardiography at follow-up
1 year prior to surgery	Heart failure—severe mitral regurgitation
Surgery	Surgical mitral and aortic valve replacement with closure of the Ao–RV fistula
2 weeks after surgery	Patient discharge

Case presentation

An 86-year-old man with an history of degenerative aortic stenosis (Supplementary material online, Video S1) and treated by TAVI (Edwards Sapiens 3, 29 mm) 3 years ago, coronary artery disease (pre-TAVI coronary angiography with a 50–70% bifurcation lesion of the anterior descending involving the first diagonal branch), a permanent bicameral pacemaker implanted 6 months ago because sick sinus syndrome and arterial hypertension. In the month following the

TAVI procedure, an Ao–RV fistula was noticed during a routine transthoracic echocardiography with a restrictive, non-significant shunt (Supplementary material online, Videos S2 and S3). No specific treatment was required and a standard pharmacological treatment with conventional clinical and echocardiography follow-up was programmed.

During the last year, the patient had worsening heart failure symptoms (progressive oedema of the lower limbs, New York Heart Association dyspnoea class III, and acute pulmonary oedema episodes) which prompted multiple hospitalizations. A new severe mitral insufficiency was diagnosed at echocardiography follow-up by his cardiologist. After case review by our hospital heart team, a surgical mitral valve replacement was decided despite the complexity of the case and high perioperative risk (EUROSCORE II 28.18%, Clinical frailty scale 4/7 ‘Vulnerable’) due to the lack of other therapeutic options offering a complete treatment. He was admitted for elective surgery and patient consent was obtained before the procedure for the academic use of his clinical data. At admission, the patient had a regular cardiac rhythm a 70 b.p.m., with a pansystolic 3/6 mitral regurgitation murmur, basal bilateral pulmonary rales, a discrete lower limb oedema, and a blood pressure of 125/56 mmHg.

The preoperative transthoracic echocardiography confirmed a degenerative and calcified mitral valve with a posterior jet of severe regurgitation of mixed aetiology mainly due to tethering of P2

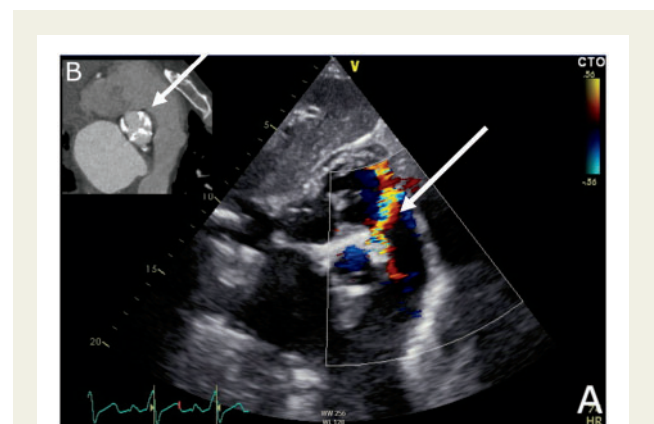


Figure 1 (A) Transthoracic colour Doppler echocardiography, short-axis view of the aorta–right ventricular fistula in telediastole (arrow). (B) Pre-transcatheter aortic valve implantation computed tomography scan (in the same orientation of the main image) showing calcifications in the base of the right coronary leaflet of the stenotic aortic valve.

secondary to left ventricular remodelling (Carpentier classification IIIb; [Supplementary material online, Video S4](#)), a dilated left ventricle (telediastolic volume index of 97 mL/m²) with an ejection fraction of 46%, anterior wall hypokinesia ([Supplementary material online, Video S5](#)), a dilated left atrium (55 mL/m²), and a systolic pulmonary artery pressure estimated at 45 mmHg. The TAVI valve had a mean gradient of 6 mmHg and a surface of 2.2 cm². An image consistent with an Ao-RV fistula, with restrictive continuous flow (V_{\max} 4.2 m/s) was noticed between the anterior aspect of the aortic valvular annulus, immedi-

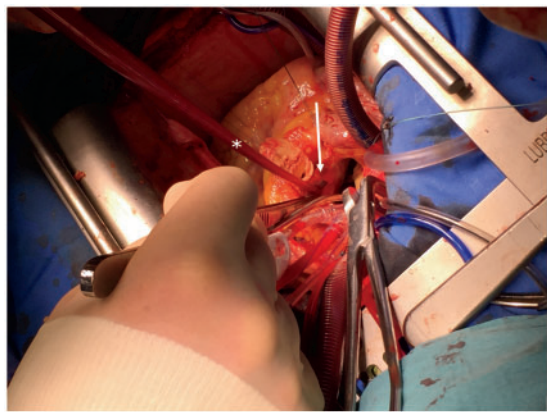


Figure 2 Surgical view of the aorta–right ventricular fistula (white arrow) after transcatheter aortic valve implantation and aortic root explantation, with the plastic aspiration probe (white *) passing through the fistula.

ately distal to the valvular plan of the aortic prosthesis, and the right ventricular side of the interventricular septum, adjacent to crista supraventricularis with an anterior directed jet, this was coincident with annular calcifications in the base of the right coronary leaflet of the aortic valve, prior to TAVI ([Figure 1, Supplementary material online, Videos S6 and S7](#)). A preoperative coronary angiography confirmed a severe stenosis (70–90%) of the proximal left anterior descending coronary artery (LAD) prompting the indication for coronary bypass. The mitral valve anatomy was considered not suitable for percutaneous edge to edge mitral valve repair because of severe annular calcification. The echocardiographic findings were confirmed by the intraoperative transoesophageal echocardiography ([Supplementary material online, Videos S8 and S9](#)) and direct surgical exploration of the diseased valve.

The patient underwent cardiac surgery under standard cardiopulmonary bypass (total time 156 min; aortic cross-clamp 138 min) with explantation of the TAVI (to better expose the mitral valve for replacement and the Ao-RV fistula for closure), surgical replacement of the mitral valve with a porcine stented bioprosthetic valve (St Jude Epic, size 31), coronary bypass (left internal thoracic artery to LAD), direct closure of the Ao-RV fistula ([Figure 2](#)), and total aortic root replacement using a porcine stentless bioprosthetic valved aortic root (Medtronic Freestyle, size 27) because of anatomic alteration of the aortic root after TAVI explantation.

The patient had a standard post-operative recuperation and was transferred to a cardiac rehabilitation facility 2 weeks after surgery. Post-operative echocardiography confirmed the absence of Ao-RV and the normal function of both implanted valves ([Supplementary material online, Video S10](#)). The patient was followed in our outpatient clinic and is still doing well 2 months after the procedure.

Table 1 Reported cases

Author	Age/sex	TAVI type	Diagnostic delay	Qp/Qs	Symptoms	Treatment	Outcome
Pilgrim 2010	91/female	Edwards Sapien	4 weeks	1.4	Exertional dyspnoea	Coil closure	Small residual shunt, no symptoms
Munoz-Garcia 2011	85/male	CoreValve (post-dilatation)	3 days	1.53	No	No	Asymptomatic at 1 year
Shakoor 2015	89/male	Edwards Sapien	1 day	1.39	Worsening HF	Conservative	Died at follow-up
Leroux 2016	47/male	Edwards Sapien	3 weeks	NA	Dyspnoea	Medical	Died, Sudden at ~4 months
Leu 2016	78/female	Edwards Sapien	2 weeks	1.13	No	No	Asymptomatic at 4 years
Almanfi 2017	92/male	Edwards Sapien XT	1 day	NA	No	No	Asymptomatic at 1 year
Nakamura 2017	54/male	Edwards Sapien XT	5 days	1.7	Cardiogenic shock	Septal Occluder	Discharged asymptomatic
Hagiwara 2017	83/male	Edwards Sapien XT	7 days	1	Mild haemolytic anaemia	Medical	Asymptomatic at 9 months
Konda 2017	88/female	Edwards Sapien XT	4 days	2	Worsening HF after 3 months	Conservative	Died at 9 months
Current case	86/male	Edwards Sapiens 3	1 month	1	HF due to concurrent mitral regurgitation	Direct surgical closure ^a	Asymptomatic 2 months after surgery

Patients from refs^{5–13} and current case.

HF, heart failure; NA, non-available; RV, right ventricular.

^aSurgical indication was related to the severe mitral regurgitation.

Discussion

This complex case highlights that cardiac surgery should not be excluded based only in the operative risk if no other therapeutic options are available and a significant clinical benefit is expected. Perioperative risk scores, such as EUROSCORE II, tend to overestimate mortality in high-risk patients⁴ and including frailty evaluation may improve outcome prediction.⁵ A tailored approach should prevent to inaccurately deny surgery to elderly patients.

Aorta–right ventricular fistula is a rare entity. Most cases reported are due to a rupture of a congenital coronary sinus aneurysm. Other aetiologies include endocarditis, trauma, and iatrogenic Ao–RV fistula.⁶ The latter is described after cardiac surgery in aortic valve or aortic root replacement and after TAVI.

Aorta–right ventricular fistula is an extremely infrequent complication after TAVI, being a subtype of aortic annular rupture. With more than 400 000 TAVI performed worldwide since the beginning of the technique this is only the 10th reported case in the literature (Table 1).^{7–15} Including our case, nine of the 10 reported cases developed the fistula after implantation of an Edwards Sapiens TAVI, probably related to balloon dilatation in a non-decalcified aortic valve during the implantation procedure. The last case had a post-dilatation procedure after Medtronic CoreValve TAVI implantation.¹¹

All of the reported cases were of late-onset (24 h to 1 month), which suggests that the fistula is a secondary complication, probably after aortic annulus haematoma.

Three patients died at follow-up (two from heart failure and one from sudden cardiac death, respectively). Of the surviving patients reported, four were not treated (reported Qp/Qs ranging from 1 to 1.53), one patient had the fistula closed with an atrial septal occluder and one with coil embolization.

Our patient is the first reported case treated surgically, but his surgical indication was related to the severe mitral regurgitation and not directly related to the Ao–RV fistula. Despite the fact that the Ao–RV fistula was not haemodynamically significant in our patient, its closure was feasible without a great increase in the surgical complexity of the case, avoiding the possible progression to a significant fistula with difficult therapeutic options. Along with the balloon post-dilatation, the absence of calcium debridement may contribute to the formation of the fistula.

Conclusion

Complex cardiac surgery should be proposed to patients who might benefit despite the operative risk if no therapeutic options are available and a net clinical benefit is expected. Despite the rarity of Ao–RV fistula after TAVI it must be considered, especially in the echocardiography follow-up of the patients. Surgical replacement of the prosthesis and closure of the fistula is a therapeutic option for this entity. In highly calcified aortic valves, the benefits of surgical debridement must be considered in the therapeutic decision-making.

Lead author biography



Between 2000 and 2011 Mario Verdugo-Marchese obtained his medical degree and internal medicine speciality in Valparaiso, Chile. From 2012 to 2017, he completed a cardiology specialist formation in the Virgen de las Nieves Hospital in Granada, Spain. Between 2017 and 2018, he worked in clinical and pre-clinical research in cardiac echocardiography in Louis Pradel Hospital in Lyon, France. Since 2018, he is working in the Cardiac Surgery Service of the Lausanne University Hospital, in Lausanne, Switzerland.

Supplementary material

Supplementary material is available at *European Heart Journal - Case Reports* online.

Slide sets: A fully edited slide set detailing this case and suitable for local presentation is available online as [Supplementary data](#).

Consent: The author/s confirm that written consent for submission and publication of this case report including image(s) and associated text has been obtained from the patient in line with COPE guidance.

Conflict of interest: none declared.

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