Iatrogenic Aorto–Right Ventricular Fistula: A Rare Complication of Transcatheter Valve Implantation

Bianca Coelho, Bsc, DMU, Martin Ng, MBBS, PhD, Chris Naoum, Bsc, MBBS, PhD, Mina Banoub, Bsc, DMU, Isabelle Saad, Bsc, and Amanda Salama, Bsc, DMU, *Macquarie Park, New South Wales, Australia*

INTRODUCTION

Transcatheter aortic valve implantation (TAVI) is a well-established therapeutic alternative to surgical aortic valve replacement (SAVR).[1] In particular, high operative risk patients who present with symptomatic severe aortic stenosis (AS) have been recommended for TAVI.[2] Conventional aortic valve replacement surgery has a higher all-cause mortality than TAVI.[3] Although TAVI is a successful procedure overall, there are still rare but serious complications that can arise. Approximately 0.2%-1.1% of cases have resulted in complications specifically involving the aorta, aortic valve (AV) annulus, or left ventricle (LV).[4] Careful procedural planning and execution is mandated due to the high mortality rate associated with annular rupture.[3] This case study discusses the implications of TAVI, specifically the formation of an aorto–right ventricular (ARV) fistula post-procedure. We present the current knowledge on prevention, diagnosis, and management of this complication.

Case Presentation

A symptomatic 89-year-old man with a body surface area of 1.97 m^2 presented with worsening dyspnea on exertion. They denied any angina, presyncope, or syncope. Comorbidities of this patient included a history of gastrointestinal bleeding, subsequent anemia, lung adenocarcinoma treated with chemoradiotherapy, and chronic atrial fibrillation treated with anticoagulation. A transthoracic echocardiogram (TTE) showed a normal size LV with mild left ventricular (LV) hypertrophy (LV indexed mass = 118 g/m^2) and normal LV systolic function (LV ejection fraction by biplane method of disks: 57%). The AV was severely calcified with a peak velocity of 4.1 m/sec, a peak pressure gradient of 68 mm Hg, and a mean pressure gradient of 42 mm Hg. The valve area was 0.87 cm^2, and the dimensionless severity index was 0.23. Doppler measurements were suggestive of severe AS. The right ventricle (RV) was dilated (RV basal diameter = 5.1 cm) with reduced right ventricular (RV) function (tricuspid annular plane systolic excursion = 1.6 cm; RV S′ = 8 cm/sec) and increased RV systolic pressure (RVSP) measured at 48 mm Hg. The patient was evaluated by the cardiothoracic team and deemed high risk for SAVR based on their advanced age and comorbidities.

Coronary angiography showed a right dominant circulation with minor coronary disease. Multidetector computed tomography (MDCT) for TAVI showed large nodules of annular calcification at the nadirs of all 3 cusps that extend onto the aortomitral continuity beneath the right coronary cusp but only protrude mildly into the LV outflow tract (LVOT) space. This patient was recommended for TAVI and underwent implantation of a balloon expandable Edwards SAPIEN 3 29-mm valve via transfemoral access. The valve was deployed in a pressure-limited manner to 5 atmospheres with deployment balloon underfilled by 2 mL along with rapid ventricular pacing at 200 bpm. A single postdilatation was then performed. The case was deemed low risk and was performed under conscious sedation with no intraoperative transesophageal echo (TEE).

Post–balloon expansion contrast aortogram showed no paravalvular leak (PVL) and no communication between the aorta and the adjacent chambers (Figure 1, Video 1).

The contrast aortogram image acquired post-TAVI shows insufficient opacification. This could be further improved by increasing the volume of contrast delivered while increasing the flow rate at a fixed contrast injection duration to obtain a higher magnitude of enhancement.

A postimplant TTE showed a well-seated valve with no PVL and a continuous shunt from the aorta (right sinus) to the RV. A velocity of 2.9 m/sec and peak pressure gradient of 34 mm Hg were measured across the shunt using continuous-wave Doppler (Figure 2). The defect measured 0.87 cm from the apical 5-chamber view and 0.45 cm from the parasternal long-axis view (Figure 3-5, Videos 2-4). The LV function was preserved (LV ejection fraction by biplane method of disks = 54%) with a dilated RV (RV basal diameter = 4.9 cm) and reduced RV function (tricuspid annular plane systolic excursion = 1.3 cm; RV S′ = 7 cm/sec). The RVSP was raised at 58 mm Hg. The ratio of pulmonic to systemic blood flow (Qp:Qs) was measured at 1.14.

The patient was referred for a TEE and a computed tomography (CT). The CT (Figure 6) showed a disruption of the right coronary sinus with communication into the RV. The maximum diameter of the opening was 0.6 cm (Figure 6). Figure 7 shows a three-dimensional reconstruction of the fistula.

From the MQ Health Cardiology, Macquarie University Hospital, Macquarie Park, New South Wales, Australia.

Keywords: Transcatheter aortic valve implantation, Aorto–right ventricular fistula, Computed tomography, Transesophageal echocardiography, Transthoracic echocardiography

Correspondence: Bianca Coelho, Bsc, DMU, MQ Health Cardiology, 2 Technology Place, Macquarie University Hospital, Macquarie Park, New South Wales 2109, Australia. (E-mail: *bianca.coelho@mqhealth.org.au*).

2468-6441
https://doi.org/10.1016/j.case.2023.01.002

VIDEO HIGHLIGHTS

Video 1: Contrast-enhanced aortogram PBD of the transcatheter AV. There was no evidence of PVL or communication between the aorta and the adjacent chambers, but image quality was not optimal.
Video 2: Two-dimensional TTE parasternal long-axis view without *(left)* and with *(right)* color flow Doppler demonstrates a communication between the aorta and the RV with left-to-right shunt throughout the cardiac cycle.
Video 3: Two-dimensional TTE parasternal short-axis view with color flow Doppler demonstrates the ARV fistula with continuous flow.
Video 4: Two-dimensional TTE zoomed apical 5-chamber view with color flow Doppler demonstrates the ARV fistula with continuous flow.
Video 5: Two-dimensional TEE zoomed midesophageal long-axis (142°) view without *(left)* and with *(right)* color flow Doppler demonstrates a communication between the aorta and the RV with left-to-right shunt throughout the cardiac cycle.
Video 6: Three-dimensional TEE, midesophageal volume-rendered image with color flow Doppler demonstrates the ARV fistula.
Video 7: Two-dimensional TEE, midesophageal x-plane views (35° and 121°) demonstrate simultaneous orthogonal images of the ARV fistula and the TAVI.
Video 8: Two-dimensional TTE parasternal long-axis view with color flow Doppler demonstrates a PVL in another patient post-TAVI for comparison. In this example, the flow is highly pulsatile (diastolic) and noncontinuous, in contrast to the ARV fistula, which is less pulsatile and continuous.
Video 9: Two-dimensional TTE parasternal short-axis view with color flow Doppler demonstrates a PVL in another patient post-TAVI for comparison. In this example, the flow is highly pulsatile (diastolic) and noncontinuous, in contrast to the ARV fistula, which is less pulsatile and continuous.

View the video content online at www.cvcasejournal.com.

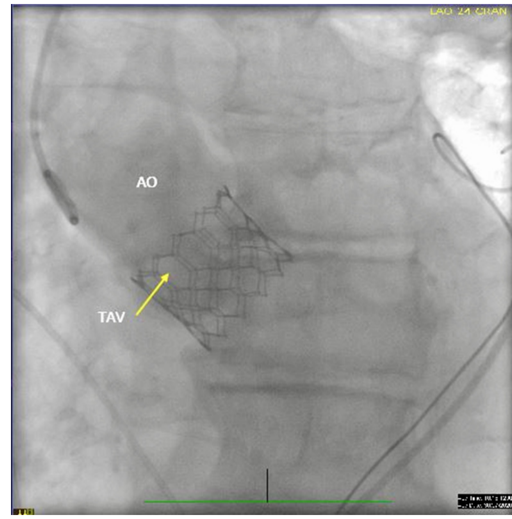

Figure 1 Contrast-enhanced aortogram (left anterior oblique, 35°; cranial, 5°) was low quality but demonstrated normal flow across the transcatheter valve (TAV; *arrow*) and no evidence of PVL or communication between the aorta and the adjacent chambers. *AO*, Aorta; *TAV*, transcatheter AV.

A TEE confirmed the presence of the communication from the aorta to the RV from the aortic root level above the AV (Figures 8-10; Videos 5-7). Flow across the fistula was continuous.

In our case, it was decided that the most likely cause of ARV fistula was the presence of heavy calcification near the right coronary cusp, which correlates with the site of the fistula (Figures 11 and 12).

A multidisciplinary team assessed the possibility of the shunt becoming hemodynamically unstable and potentially requiring intervention. However, it was deemed that the fistula tract was small, serpentine, and surrounded by calcium, making it difficult to traverse with a percutaneous closure device and therefore unsuitable for percutaneous closure.

The patient was discharged 5 days post TAVI, as they were asymptomatic and clinically well. They were managed conservatively with 3 monthly follow-up appointments to assess the hemodynamic significance of the ARV fistula and to assess for any signs of right heart failure.

At their 3-month follow-up, there was no change in the ARV fistula. The patient did develop some signs of exacerbation of heart failure, which was successfully managed by escalation in their diuretic therapy. Overall, they reported feeling less short of breath than prior to the TAVI.

The velocity across the defect remained consistent in comparison to the previous study and was measured at 2.7 m/sec; the Qp:Qs was 1.04. There was no change in their RV dimensions and LV size, and contractility was within normal limits. The RVSP remained stable at 59 mm Hg.

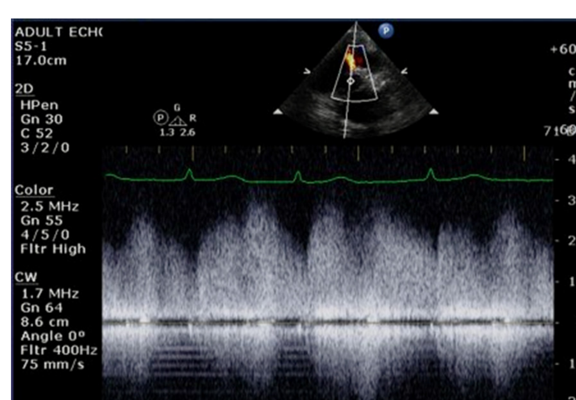

Figure 2 Two-dimensional TTE-guided continuous-wave spectral Doppler display from the parasternal short-axis view demonstrates flow across the shunt that was parallel to the ultrasound beam and ideal for estimation of the maximum velocity. There is continuous flow above the baseline suggestive of flow from the aorta to the RV with a peak velocity measured at 2.9 m/sec.

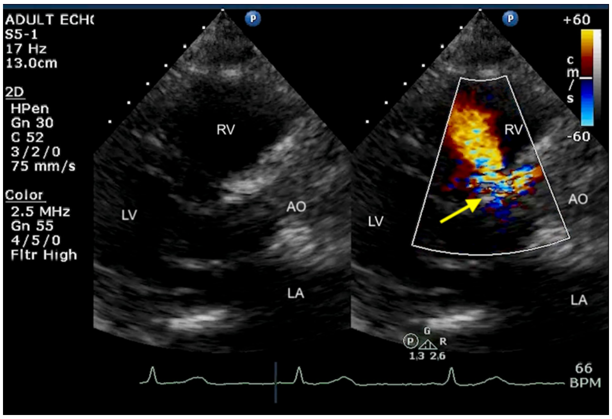

Figure 3 Two-dimensional TTE parasternal long-axis view without *(left)* and with *(right)* color flow Doppler demonstrates the ARV fistula *(arrow)* with flow from the AO to the RV. The fistula measures 0.45 cm from this view. *AO*, Aorta; *LA*, left atrium.

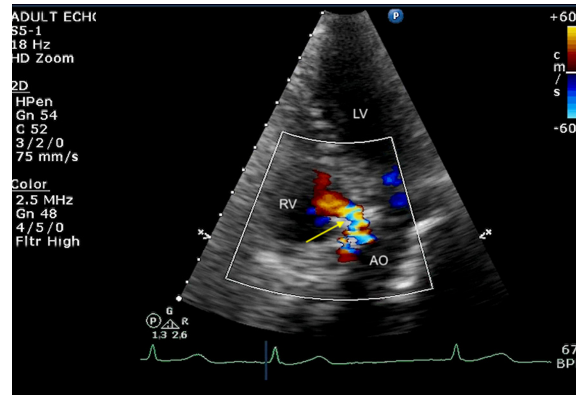

Figure 5 Two-dimensional TTE apical 5-chamber view with color flow Doppler demonstrates the ARV fistula *(arrow)*, with flow from the AO to the RV. The fistula measured 0.87 cm in this view. *AO*, Aorta.

At 8 months post-TAVI, the patient reported no worsening of breathlessness and RV dimensions remained stable. Taking this into account, along with the risks of sternotomy to repair of the fistula, the structural heart team concurred on managing this patient expectantly.

DISCUSSION

In the Western world, AS is the most common form of valvular heart disease.[5] It has been shown to occur with advancing age and is present in about 5% of the population by the age of 65. A meta-analysis of studies from Europe, United States, and Asia showed a prevalence of 12.4% of AS overall and a prevalence of 3.4% in patients 75 years and older.[6]

Techniques in TAVI are emerging, and recent trials have shown that it is fast becoming the standard of care in certain patients and an alternative to SAVR in high-risk surgical patients.[7] The progressive increase in the number of transcatheter valve implants has also increased the likelihood of rare complications occurring.[8]

There are 2 major types of transcatheter heart valves (THVs), balloon-expandable valves (BEVs) and self-expanding valves (SEVs). The main differentiating features of these valves are valve height, implantation depth, relative position of the valve and the annulus, radial force, deployment mechanism, and valve geometry. The longer stent in SEVs poses a higher risk in developing conduction abnormalities due to the anatomic proximity of the conduction system to the distal landing zone of the THV, making it vulnerable to injury during TAVI.[9]

The feasibility of coronary access for future procedures and the possibility of coronary obstruction are other important factors to keep in mind with the longer stents of SEVs.[10]

Valve selection is dependent on patient-specific anatomy, for example, coronary height and dimensions of peripheral vasculature. Self-expanding valves have been shown to result in significantly better postprocedural effective orifice areas and may be beneficial in patients with previous surgical valves and those with smaller annulus diameters, who are at high risk of patient-prosthesis mismatch. Patients

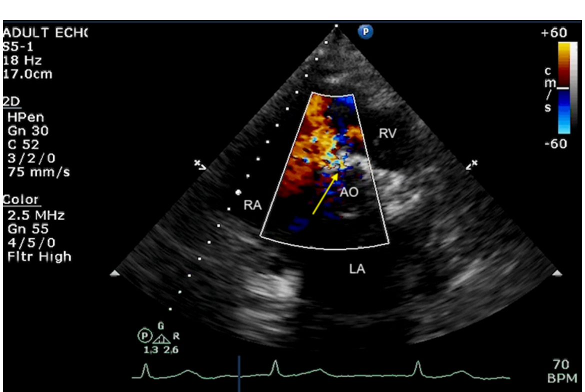

Figure 4 Two-dimensional TTE parasternal short-axis view with color flow Doppler demonstrates the ARV fistula *(arrow)* with flow from the AO to the RV. *AO*, Aorta; *LA*, left atrium.

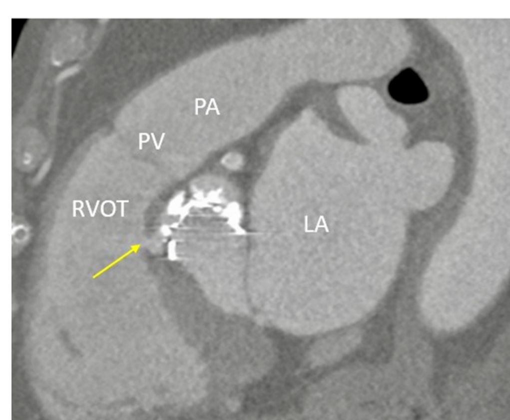

Figure 6 MDCT sagittal view demonstrates the TAVI and the location of the ARV fistula *(arrow)*. *LA*, Left atrium; *LV*, left ventricle; *PA*, pulmonary artery; *RVOT*, RV outflow tract.

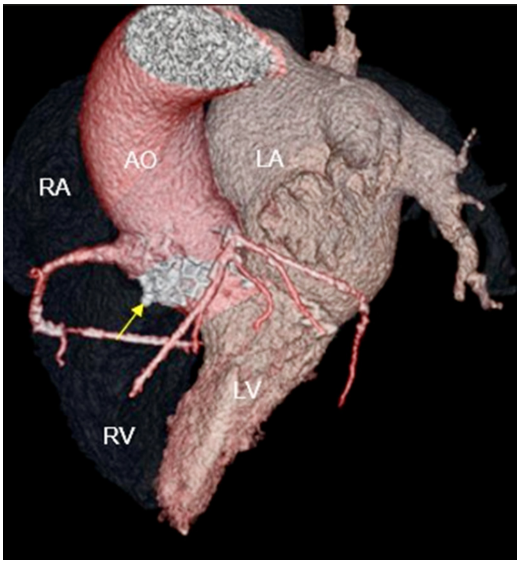

Figure 7 MDCT image, volume-rendered whole-heart reconstruction image post-TAVI demonstrates the location of the fistula *(arrow)*. The RV and RA have been faded out *(dark gray)* to better visualize the fistula. *AO*, Aorta; *LA*, left atrium; *RA*, right atrium.

that are prone to encounter postprocedural pacemaker implantation or PVL, including patients who are older and those who have prior conduction disturbances, are better suited to BEVs.[11]

The protocol for sedation in TAVI is dependent on the surgical risk of the patient and their underlying comorbidities. Conscious sedation has a significantly lower postoperative risk and shorter intensive care unit length of stay. It is suggested that limiting the processes involved in general anesthesia such as induction, orotracheal intubation, and positive pressure ventilation can lead to better hemodynamic stability and minimize the use of vasopressors and inotropes post operatively.[12]

Complications involving the aorta, AV annulus, or LV are rare. Aortic dissection after TAVI is rare, occurring at the rate of <2%, and can involve any segment of the aorta.

The interaction of the stiff wire within the ascending aorta, intimal disruption to the aortic wall by catheter valve injury, valve retraction to expose the balloons in BEVs, injury postballoon valvuloplasty, or post–balloon dilation (PBD) interaction with the aorta are all potential mechanisms in which aortic dissection may occur.[4]

Our case illustrates a rare complication of an ARV fistula that did not require repair. Although the patient's valve was sized appropriately, it was agreed that displacement of the heavily calcified valve may be the etiology for the fistula.

Studies have shown that the presence of subannular calcification increases the risk of patients developing annular rupture postimplant. It has been shown that there is an 11-fold increased risk that the balloon-expandable THV will rupture in the presence of moderate to severe LVOT/subannular calcification.[13]

There have been a few different reported mechanisms for aortic root rupture. Konda *et al.*[2] describes the presence of congenital aneurysms of the sinuses of Valsalva as being the most common cause of ARV fistulas. Coughlan *et al.*[3] reports that a high burden of LVOT calcification that extends into the LVOT and potential valve area oversizing >20% on MDCT are some of the main predictors of aortic root injury during TAVI with BEVs. Figure 13 shows potential locations for aortic annular rupture.

While the exact mechanism of fistula formation is unknown, some important factors that can present a risk for aortic annulus rupture or

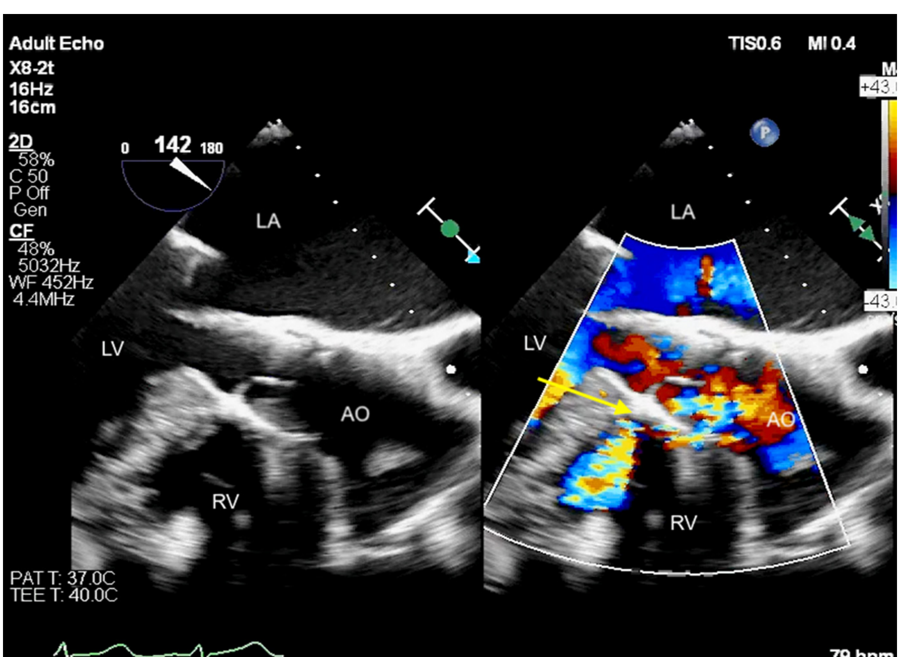

Figure 8 Two-dimensional TEE, zoomed midesophageal long-axis (142°) view without *(left)* and with *(right)* color flow Doppler demonstrates a communication between the aorta and the RV with left-to-right shunt *(arrow)*. *AO*, Aorta; *LA*, left atrium.

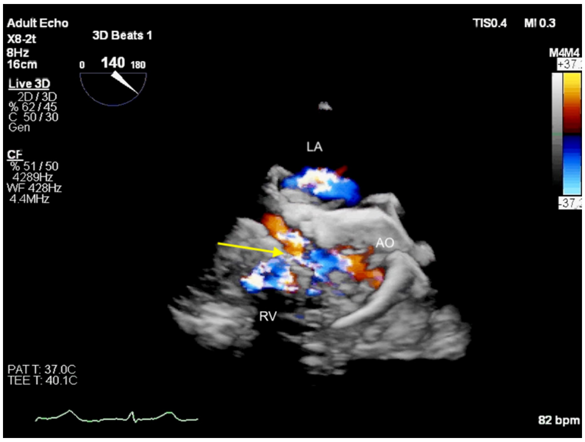

Figure 9 Three-dimensional TEE, midesophageal volume-rendered image with color flow Doppler demonstrates the ARV fistula *(arrow)*. *AO*, Aorta; *LA*, left atrium.

fistula formation mainly in the case of BEVs are the depth of prosthesis, larger size devices, and excessive postdilatation.[7,8]

Compared with SAVR, THVs are self-anchoring. The surrounding calcium together with the radial force of the valve holds the device in position. Implanters aim for 5%-10% oversizing of the THV relative to the annular diameter of the native valve, to reduce the risk of PVLs, embolization, or device migration. Post–balloon dilation after deployment of the BEV is commonly performed to reduce the presence of PVLs. However, the risk versus benefit of PBD should be taken into consideration. One recommendation is to perform PBD in all cases of grade III-IV PVLs and to consider the risks and benefits of PBD in the context of grade II PVLs.[14] Aggressive approaches to oversizing of the valve >15% of the annular diameter and PBD for minor PVLs would increase the risk of aortic root injury.

From an imaging perspective the flow within the fistula can be mistaken for a PVL in some TTE views. Figure 14 shows a comparative image of a PVL (panels A and B) and an ARV fistula (panels C and D).

Paravalvular leaks are seen in diastole with aliasing of blood flow noted within the aortic root in all views. In comparison, flow across the fistula is noted throughout the cardiac cycle and is seen moving across toward the RV. In the parasternal short-axis view, the appearance of an ARV fistula can be similar to that of PVL as shown below (Figure 14B and D). However, inferior panning of the ultrasound beam toward the RV shows the flow going across the aortic root into the RV in the case of ARV fistulas (Videos 2 and 3) and flow contained within the aortic root in PVLs (Videos 8 and 9).

The hemodynamic significance of the shunt determines the symptomatology. Small shunts with a QP:QS < 2 can be managed conservatively, while larger shunts with elevated RVSP and signs of right heart failure due to volume and pressure overload might be an indication for a repair.

In cases of aortic root rupture or fistulas, surgical repair and percutaneous device closures are treatments of choice.[2] However, the implantation of a percutaneous closure device in the case of a TAVI-related ARV fistula presents a technical challenge. The variations in size, shape, and complexity of the defect and the possibility that the closure device could interfere with the TAVI are factors that need to be considered. Nevertheless, in patients that are high-risk surgical candidates, percutaneous intervention under TEE guidance is a valuable alternative option.[8]

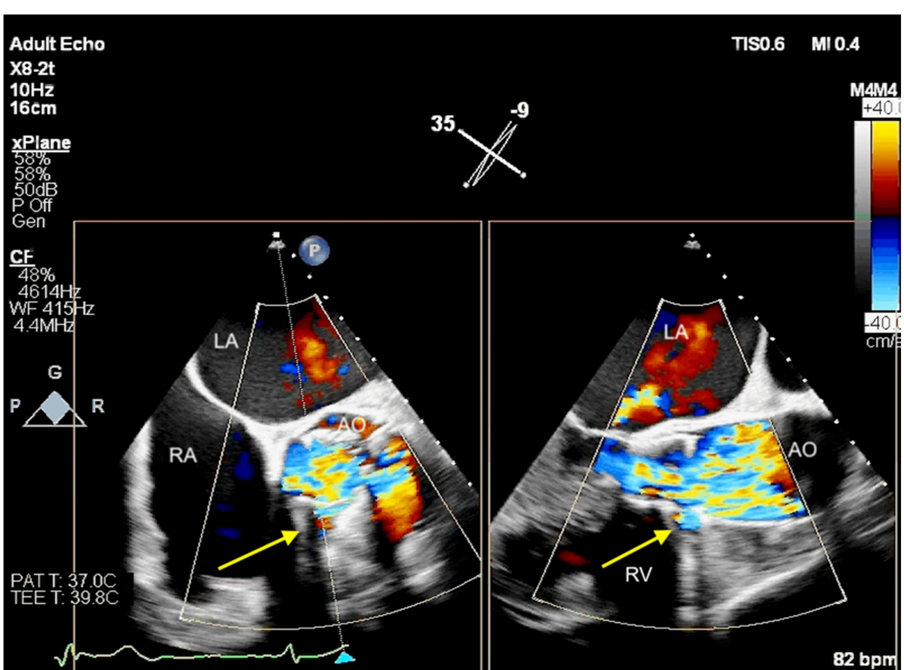

Figure 10 Two-dimensional TEE, midesophageal x-plane views (35° and 121°) demonstrate simultaneous orthogonal images of the origin of the ARV fistula *(arrows)*. *AO*, Aorta; *LA*, left atrium; *RA*, right atrium.

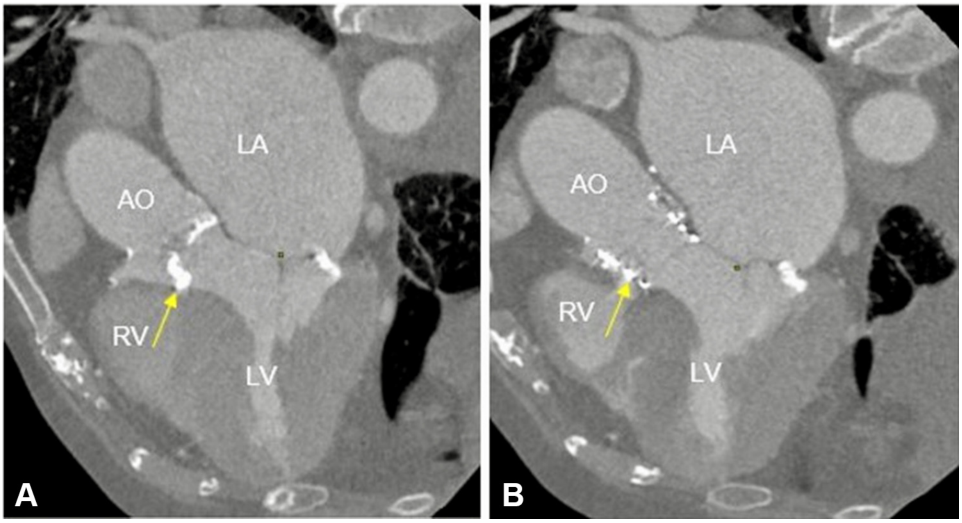

Figure 11 MDCT image, oblique axial display at baseline pre-TAVI **(A)** and post-TAVI **(B)** demonstrates focal heavy calcification within all cusps, but greatest at the right cusp *(arrow)*, which is also the location of the subsequent ARV fistula post-TAVI *(arrow)*. *AO*, Aorta; *LA*, left atrium

CONCLUSION

Aortic root rupture and formation of fistulas are a rare but serious complication of TAVI. It is essential that patients undergo preprocedure MDCT screening to assess the predictors of annular rupture, specifically subannular calcification, and to ensure that the selected transcatheter valve is appropriately sized. In the case of patients with significant subannular calcification, aggressive dilatation of the balloon must be avoided.[2] The treatment and management of patients that develop an ARV fistula are determined by the severity of flow and patient symptoms. There are no reported cases of spontaneous closure of an ARV fistula post-TAVI. In patients that are asymptomatic with hemodynamically stable shunts and no significant increase in RVSP or exacerbation of right heart failure, annual clinical reviews with supporting echo imaging are essential. The first line of treatment is surgical repair; however, percutaneous closure procedures have been reported in high surgical risk patients.[15]

ETHICS STATEMENT

The authors declare that the work described has been carried out in accordance with the following guidelines: National Statement on Ethical Conduct in Human Research from the National Health and Medical Research Council.

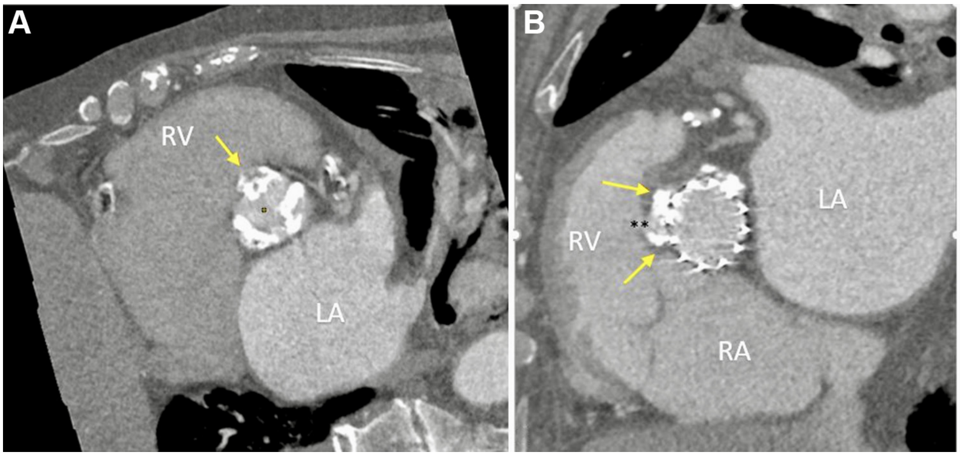

Figure 12 MDCT image, short-axis view of the aortic annulus at baseline pre-TAVI **(A)** and post-TAVI **(B)** demonstrates heavy calcification within all cusps including the right cusp *(arrow)* and its relationship to the site of subsequent ARV fistula (**). *LA*, Left atrium; *RA*, right atrium.

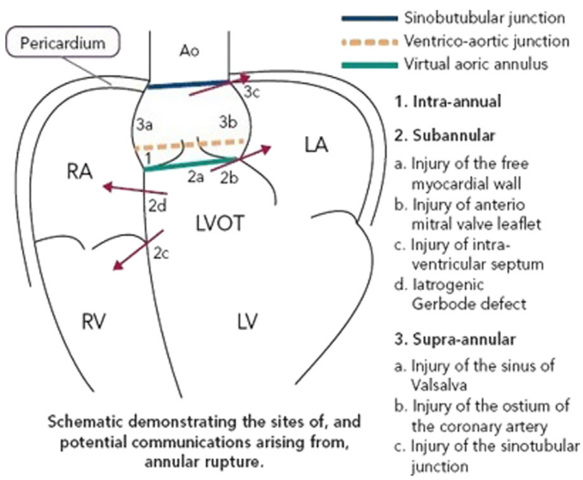

Figure 13 Potential locations for aortic annular rupture.[3] Used with permission of Radcliffe Cardiology from Coughlan *et al*.[3] Permission conveyed through Copyright Clearance Center. *Ao*, Aorta; *LA*, left atrium; *RA*, right atrium.

CONSENT STATEMENT

The authors declare that since this was a non-interventional, retrospective, observational study utilizing de-identified data, informed consent was not required from the patient under an IRB exemption status.

FUNDING STATEMENT

The authors declare that this report did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

DISCLOSURE STATEMENT

The authors report no conflict of interest.

ACKNOWLEDGMENTS

We thank Dr. Grant Shalaby (MQ Health Cardiology, Macquarie University, New South Wales) for his assistance with the contrast angiography image and description and Dr. Lisa Simmons (MQ Health Cardiology, Royal Prince Alfred Hospital, New South Wales) for all the TEE images.

SUPPLEMENTARY DATA

Supplementary data to this article can be found online at https://doi.org/10.1016/j.case.2023.01.002.

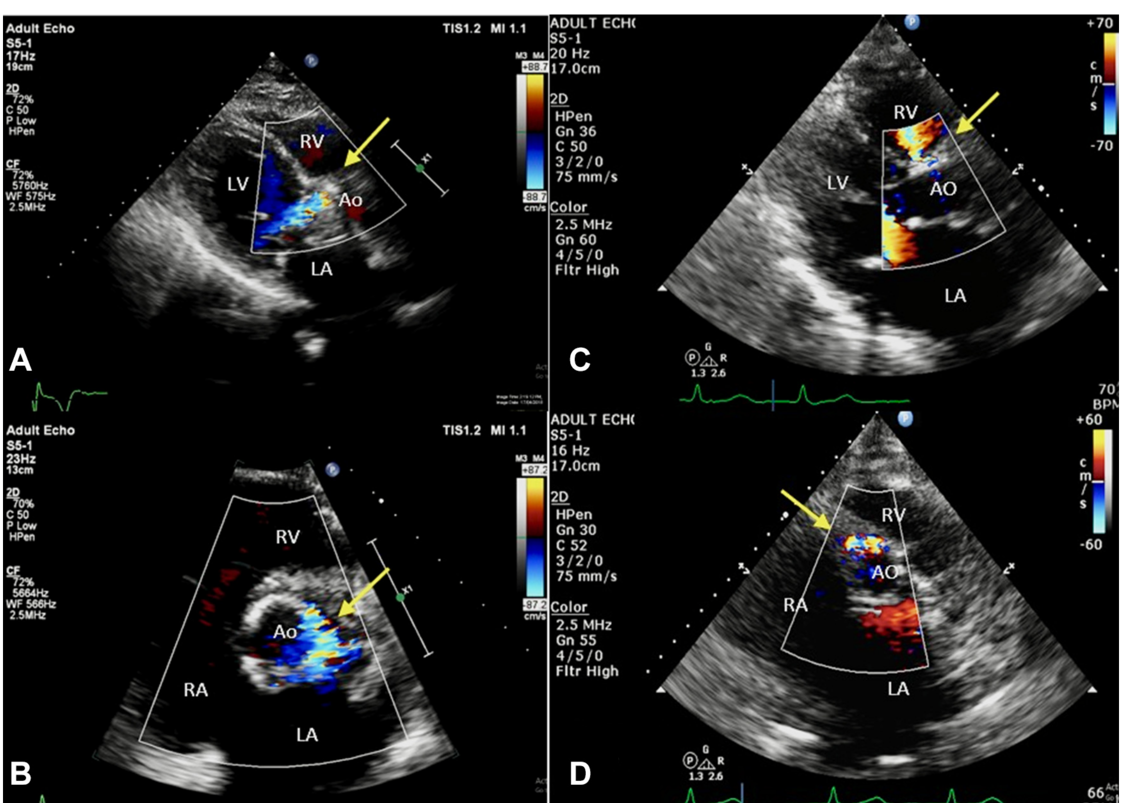

Figure 14 Two-dimensional TTE parasternal long-axis and parasternal short-axis *(bottom)* views with color flow Doppler in a patient post-TAVI with a PVL (**A, B**; *arrows*) compared to this reported patient with an ARV fistula (**C, D**; *arrows*). *Ao*, Aorta; *LA*, left atrium; *RA*, right atrium.

REFERENCES

1. Ueshima D, Fovino LN, D'Amico G, Brener SJ, Esposito G, Tarantini G. Transcatheter versus surgical aortic valve replacement in low- and intermediate-risk patients: an updated systematic review and meta-analysis. Cardiovasc Interv Ther 2019;34:216-25.
2. Konda MK, Kalavakunta JK, Pratt JW, Martin D, Gupta V. Aorto-right ventricular fistula following percutaneous transcatheter aortic valve replacement: case report and Literature review. Heart Views 2017;18:133-6.
3. Coughlan JJ, Kiernan T, Mylotte D, Arnous S. Annular rupture during transcatheter aortic valve implantation: predictors, management and outcomes. Interv Cardiol 2018;13:140-4.
4. Langer NB, Hamid NB, Nazif TM, Khalique OK, Vahl TP, White J, et al. Injuries to the aorta, aortic annulus, and left ventricle during transcatheter aortic valve replacement: management and outcomes. Circ Cardiovasc Interv 2017;10:e004735.
5. Nathaniel S, Saligram S, Innasimuthu AL. Aortic stenosis: an update. World J Cardiol 2010;2:135-9.
6. Ancona R, Pinto S. Epidemiology of Aortic Valve Stenosis (as) and of aortic valve incompetence (AI): is the prevalence of AS/AI similar in different parts of the world? [Internet]. Epidemiology of aortic valve stenosis (AS) and of aortic valve incompetence (AI): is the prevalence of AS/AI similar in different parts of the world? European Society of Cardiology. 2020 Available at: https://www.escardio.org/Journals/E-Journal-of-Cardiology-Practice/Volume-18/epidemiology-of-aortic-valve-stenosis-as-and-of-aortic-valve-incompetence-ai#:~:text=Aortic%20Stenosis,-Aortic%20stenosis%20. Accessed November 12, 2022.
7. Shakoor MT, Islam AM, Ayub S. Acquired aorto-right ventricular fistula following transcatheter aortic valve replacement. Case Rep Cardiol 2015;2015:608539.
8. Vrettos A, Duncan A, Ahmed A, Heng EL, Panoulas V. Successful percutaneous closure of aortic root-to-right ventricle fistula after transcatheter aortic valve implantation: a valuable option in high-risk surgical patients. Eur Heart J Case Rep 2022;6:ytac094.
9. Chen S, Chau KH, Nazif TM. The incidence and impact of cardiac conduction disturbances after transcatheter aortic valve replacement. Ann Cardiothorac Surg 2020;9:452-67.
10. Forrestal BJ, Case BC, Yerasi C, Shea C, Torguson R, Zhang C, et al. Risk of coronary obstruction and feasibility of coronary access after repeat transcatheter aortic valve replacement with the self-expanding evolut valve: a computed tomography simulation study. Circ Cardiovasc Interv 2020; 13:e009496.
11. Lee HA, Chou AH, Wu VC, Chen DY, Lee HF, Lee KT, et al. Balloon-expandable versus self-expanding transcatheter aortic valve replacement for bioprosthetic dysfunction: a systematic review and meta-analysis. PLoS One 2020;15:e0233894.
12. Téllez-Alarcón M, Montes FR, Hurtado P, Gutiérrez LP, Cabrales JR, Camacho J, et al. Conscious sedation versus general anesthesia for transcatheter aortic valve implantation: a retrospective study. Braz J Anesthesiol 2022;72:539-41.
13. Barbanti M, Yang TH, Rodès Cabau J, Tamburino C, Wood DA, Jilaihawi H, et al. Anatomical and procedural features associated with aortic root rupture during balloon-expandable transcatheter aortic valve replacement. Circulation 2013;128:244-53.
14. McInerney A, Vera-Urquiza R, Tirado-Conte G, Marroquin L, Jimenez-Quevedo P, Nuñez-Gil I, et al. Pre-dilation and post-dilation in transcatheter aortic valve replacement: indications, benefits and risks. Interv Cardiol 2021;16:e28.
15. Verma S, Kakar P, Moskovits N, Shani J, Frankel R. Aorta to RV fistula: a rare complication of TAVR. Cath Lab Digest 2018;26.