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CASE REPORT
Coronary heart disease

Case report: iatrogenic left ventricular outflow tract to right atrium fistula after trans-femoral transcatheter aortic valve implantation associated with asymmetric septal hypertrophy

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Background The transcatheter aortic valve implantation (TAVI) is becoming a leading treatment option for symptomatic aortic stenosis for patients in all surgical risk categories. Recognition and management of potential complications are essential to ensure patient life and comfort. We present here a case report of a left ventricular outflow tract (LVOT) to right atrium (RA) fistula which is an extremely rare complication after TAVI.

Case summary An 85-year-old man with symptomatic severe aortic stenosis and non-obstructive asymmetric septal hypertrophy (ASH) underwent a transfemoral TAVI. Soon after the procedure, he developed chest pain and atrial fibrillation with rapid ventricular response. A transthoracic echocardiography followed by a transoesophageal echocardiography showed a small pseudo-aneurysm with a fistulous tract between the LVOT and the RA. This was confirmed by a contrast computed tomography scan of the heart. The patient remained asymptomatic throughout the rest of hospitalization. He was treated with diuretics and discharged home. One month follow-up showed increase in the width, jet size, and gradient of the fistula but the patient remained asymptomatic. The decision by Heart team was to closely monitor him for symptoms since the fistula is difficult to access percutaneously.

Discussion We report a unique case of an LVOT to RA fistula in the setting of ASH that occurred post-TAVI. Alcohol septal ablation was proposed pre-TAVI for patients having septal thickening >15 mm and dynamic obstruction. Treatment options for iatrogenic fistula vary from symptomatic treatment to percutaneous or surgical closure.

Keywords LVOT to RA fistula • Post-TAVI complications • Asymmetric septal hypertrophy • Case report

Learning points

- Left ventricular outflow tract to right atrium fistula is a rare complication post-transcatheter aortic valve implantation (TAVI). Its mechanism is not well understood.
- Alcohol septal ablation has been proposed as a new treatment strategy for patients with asymmetric septal hypertrophy >15 mm and dynamic obstruction associated with aortic stenosis before TAVI.

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Introduction

Transcatheter aortic valve implantation (TAVI) has become a safe and effective procedure for severe symptomatic aortic stenosis (AS) in patients with prohibitive, high, intermediate, and low risk for surgical aortic valve replacement.[1–6] Commonly reported complications post-TAVI were paravalvular leak (PVL), permanent pacemaker implantation, cerebrovascular accident (CVA), myocardial infarction (MI), and vascular complications (VCs).[7] Aorta to the right ventricle fistula is an infrequent complication post-TAVI reported in six cases until 2018.[8] Even less frequently, left ventricular outflow tract (LVOT) to the right atrium (RA) fistula was reported once following TAVI in a patient with a heavily calcified aortic valve and annulus.[9] Here, we present a first reported case of an LVOT to the RA fistula in a patient with asymmetric septal hypertrophy (ASH).

Timeline

Day	Events
14 June 2020	Admission
15 June 2020	Aortic valve implantation via femoral route.
	Chest discomfort post-transcatheter aortic valve implantation.
	High pitched holosystolic cardiac murmur.
	Transthoracic echocardiography showing fistula.
17 June 2020	Transoesophageal echocardiography showing aorto-atrial fistula between the left ventricular outflow tract and the right atrium.
	No symptoms reported.
18 June 2020	Cardiac computed tomography performed.
22 June 2020	No symptoms reported.
	Discharge from hospital.
	Follow-up transthoracic echocardiography was prescribed.
27 July 2020	Transthoracic echocardiography (follow-up).

Case presentation

An 85-year-old man with a medical history significant for ASH, hypertension, hyperlipidaemia, and severe aortic stenosis presented to our centre for worsening dyspnoea with the New York Heart Association (NYHA) Class III symptoms with no reported angina nor syncope. On physical examination, his blood pressure was 130/80 mmHg, heart rate was 76 b.p.m. Cardiac auscultation was remarkable for an aortic systolic murmur best heard at the second intercostal space with a significant decrease in S2. The transthoracic echocardiogram (TTE) showed ASH with a diastolic septal diameter of 17 mm, a fixed LVOT gradient of 11 mmHg, moderately calcified aortic valve associated with severe stenosis (mean gradient of 42 mmHg, the maximal velocity of 4.05 m/s, the continuity equation was not applicable to calculate aortic valve area due to accelerated

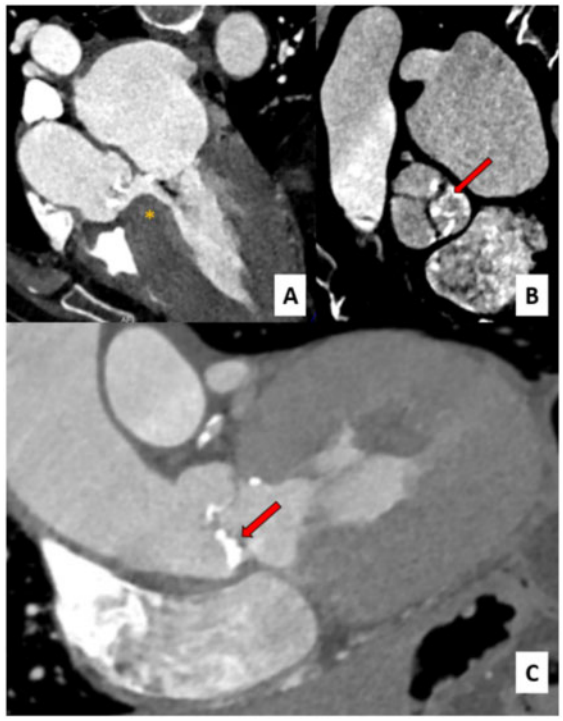


Figure 1 (*A–C*) Pre-procedure computed tomography scan showing asymmetric septal hypertrophy (orange star) and moderately calcified aortic valve (red arrow).

jet in the LVOT), an ejection fraction (EF) of 60%, and normal systolic pulmonary artery pressure. Cardiac catheterization revealed normal coronary angiogram. He had a low risk for open-heart surgery (Logistic Euro score 7.96%, STS score 2.9%) with a frail physical state for which the Heart team decided to refer him for TAVI. Computed tomography and angiography showed ASH with a moderately calcified aortic valve, mild LVOT calcifications (*Figure 1A–C*), and an accessible femoral route.

The TAVI procedure was performed with a 26 mm Sapien 3 bioprosthetic valve (Edwards Lifesciences, Irvine, CA, USA). The initial result was acceptable, with no PVL nor coronary obstruction. We noticed an upward movement of the valve during implantation (*Figure 2B*, *Video 1*). There was no pre- nor post-dilations required. No electrical or access site complications were encountered. The patient was transferred to the cardiac ward.

Later that day, the patient developed chest discomfort associated with atrial fibrillation with a rapid ventricular response at 130 b.p.m. and non-specific ST-T changes. A TTE showed normal EF, a mean aortic gradient of 8 mmHg, a fistula jet reaching from the LVOT to the RA, no right ventricular dilation, no right atrial dilation, and no pericardial effusion. Transoesophageal echocardiogram showed mild PVL, and an LVOT pseudo-aneurysm, that is associated with an LVOT to the RA fistula. The fistula's jet was lining the tricuspid valve plane with a maximal gradient of 123 mmHg (*Figure 3A–C*, *Video 2*). A post-procedure computed tomography (CT) scan showed that the

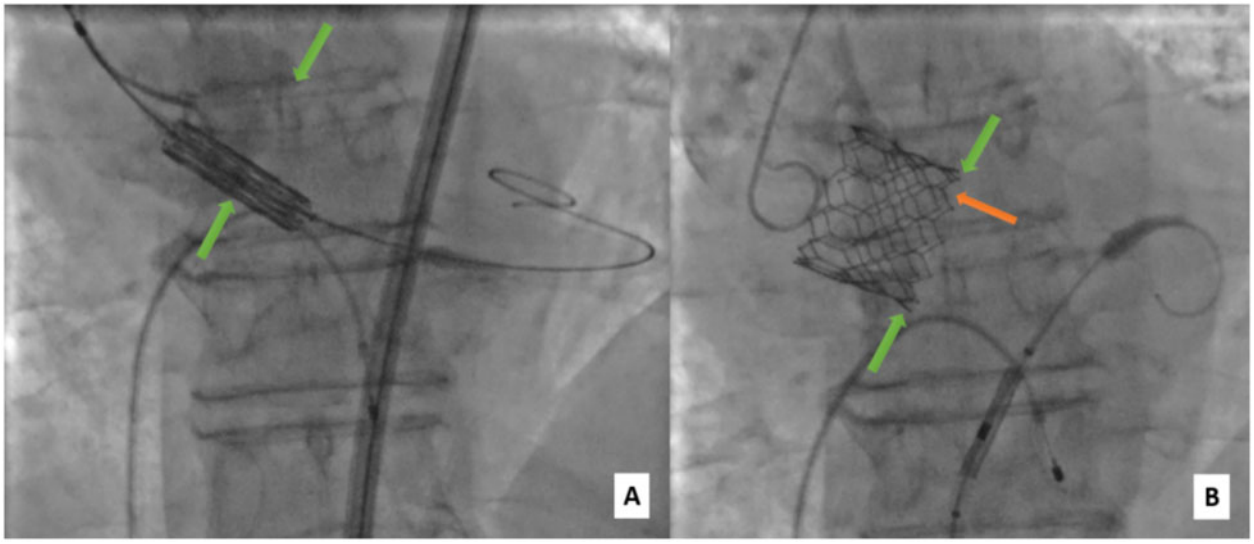
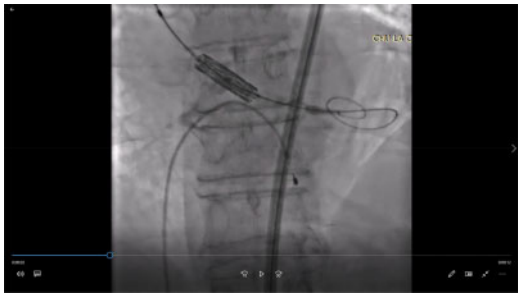


Figure 2 Angiography showing position of the bioprosthesis. (*A*) Pre-inflation. (*B*) Post-inflation. The orange arrow shows the upward movement of the valve. The green arrows show the annular plane.

bioprosthesis was well placed with no significant PVL, no coronary artery obstruction. At the level of the LVOT, there exists a small pseudo-aneurysm of 5 mm, immediately upstream of the valve prosthesis. It is extended by a fistulous track towards the RA of an ~2 mm diameter, where the jet flushes parallel to and outlining the tricuspid valve plane (*Figure 4A–D*).

The patient remained asymptomatic during the course of the hospitalization and was discharged home on Day 7 post-TAVI. The follow-up TTE was done 1 month later showed a persistence of the fistula with an increase in size (from 2 to 2.95 mm) and an increased colour jet and gradient (maximal gradient 166 mmHg) (*Figure 5A–D*, *Video 3 and* Supplementary material online, *Video S1*).



Video 1 Cine loop of valve implantation.

Discussion

The complications of TAVI, including CVAs, PVL, MIs, and VCs, are commonly described, where most of them have prognostic implications. Others, like aorto-ventricular and aorto-atrial fistula, are rare complications. Left ventricular outflow tract to RA fistula is an even rarer complication. Their mechanism is not fully understood. They are challenging to predict, and their prevention and treatment pathways are not well established, to be tailored for each case apart. Our article presented a case of iatrogenic LVOT to RA fistula post-TAVI associated with ASH. This case is the first reported in this context where the fistula's position is not usual. A case of latrogenic atrioventricular septal defect was reported by Salizzoni *et al.* in 2014,[9] in the context of severe aortic and annular calcifications.

The combination of AS and ASH has been found in 10% of the patients with AS undergoing surgery.[10] The treatment consisted of surgical septal myomectomy with surgical aortic valve replacement.[11] In patients with high surgical risk or contraindications for surgery, alcohol septal ablation (ASA) before TAVI was suggested as a potential treatment strategy by Khan *et al.*[12] in patients with ASH (>15 mm) and dynamic LVOT obstruction. In our case, the LVOT gradient was fixed at 11 mmHg, so no ASA was needed before TAVI.

The mechanism of the fistula was not fully understood. No annular calcifications were covering the area of concern. We suggest that a direct trauma by the metallic frame of the bioprosthesis was the fistula's cause, as we can see contralateral impregnation on the septal wall facing the fistula (*Figure 4C*). We believe that the presence of ASH has left no room for the bioprosthesis expansion, which caused an upward movement of the valve in the moment of inflation (*Figure 2B*, *Video 1*) that resulted in direct trauma to and the perforation of the membranous septum, as we can see in the *Figure 4D*. To prevent this type of complication, we need to avoid oversizing, choose the proper type of valve that suits the LVOT/valve complex (Bicuspid, calcified LVOT, ASH) throughout pre-planning, and to avoid post-dilations of the bioprosthesis.[13] The valve area in systolic reconstruction was measured as 571 mm^2, which corresponds to a bioprosthesis diameter of 29 mm with a 13.9%

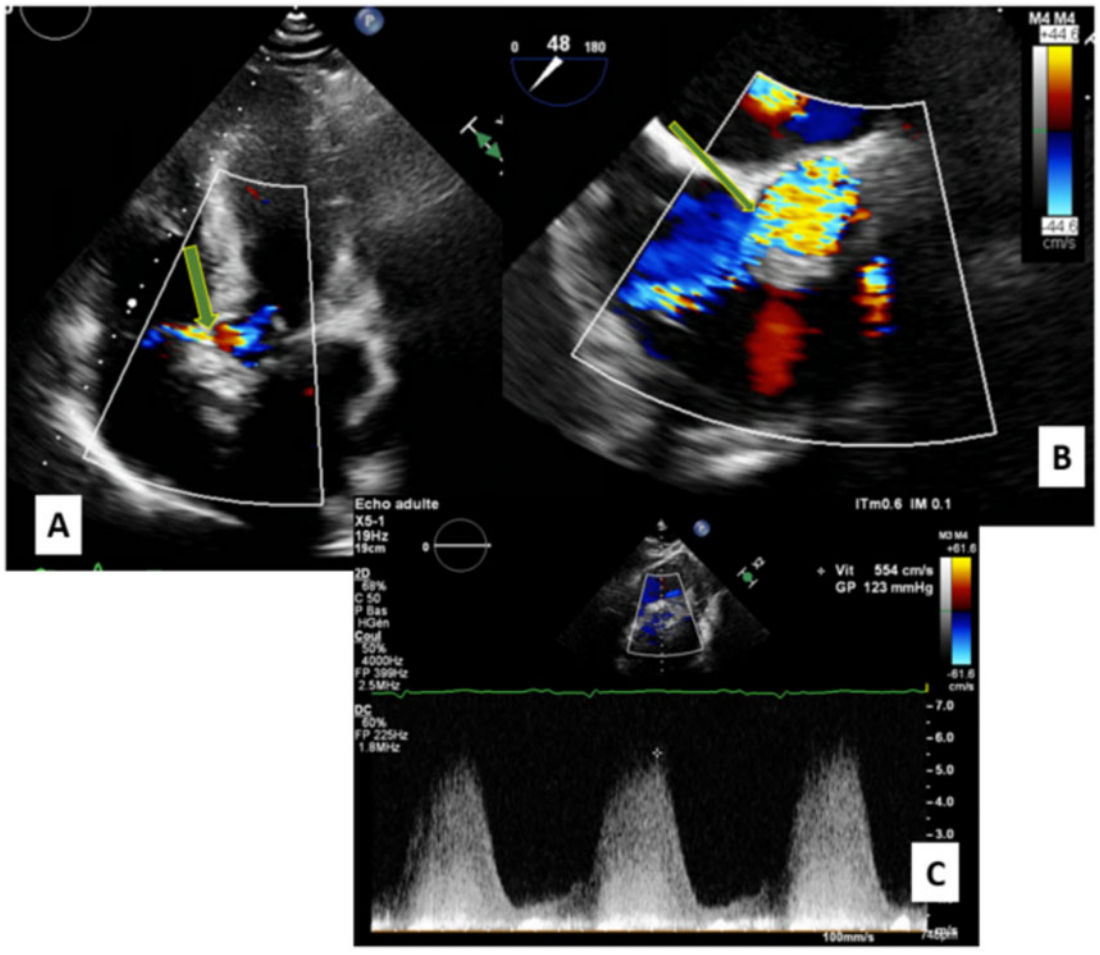
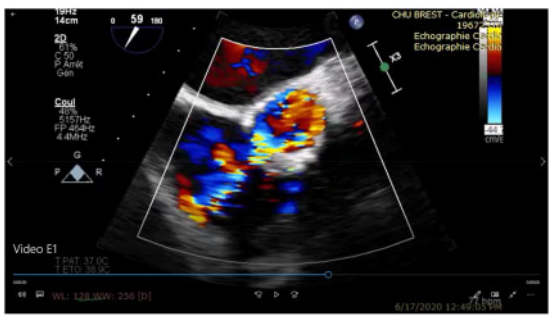
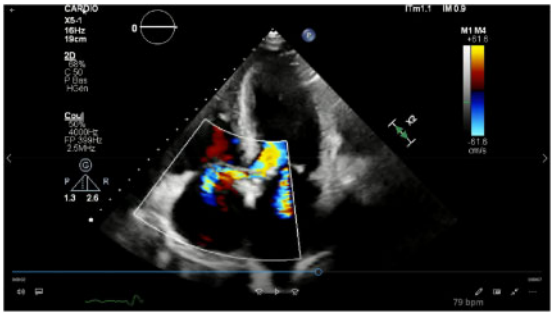


Figure 3 (*A*) Transthoracic echocardiography (apical four-chamber view) showing the left ventricular outflow tract-right atrial fistula jet lining the plane of the tricuspid valve (green arrow). (*B*) Transoesophageal echocardiography showing the pseudo-aneurysm with fistula jet (green arrow). (*C*) Continuous Doppler showing fistula maximal gradient of 123 mmHg.



Video 2 TEE color Doppler showing the pseudoaneurysm and the fistula. The jet is lining the tricuspid valve plane.



Video 3 Follow up TTE color Doppler apical view showing the fistula and the jet.

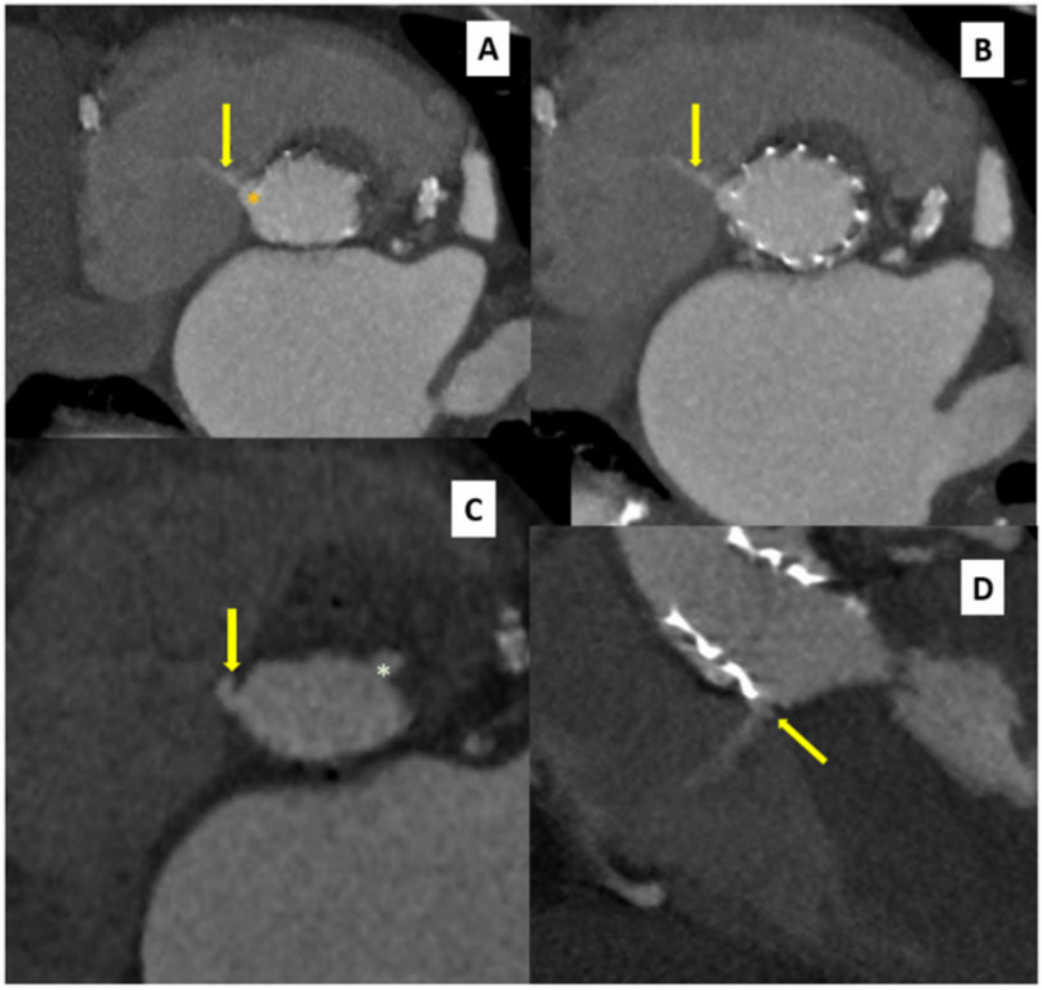


Figure 4 Post-procedure computed tomography scan. (*A–D*) showing: the left ventricular outflow tract aneurysm (yellow star), the fistula, and its jet lining the plane of the tricuspid valve (yellow arrow). (*C*) The contralateral impregnation over the intraventricular septum (white star).

oversizing estimation. We used a 26 mm bioprosthesis with an 8.9% undersizing estimation to anticipate any likely oversizing damage.

Due to fistula's rare occurrence, no randomized clinical trials will ever be designed to define this complication's best treatment option. It is generally recommended to repair fistulae via surgical or percutaneous approaches if it grades more than trivial because it is likely worsening over time. It may cause overt right heart failure and cardiac arrhythmias such as atrial fibrillation.[14] The percutaneous approach consists of deploying a small occluder device (Amplatzer Vascular Plug, AVP) to close the fistula. This modality was reported in a case by Vainrib *et al.*[15] describing the treatment of an aorto-right ventricular fistula post-TAVI. Small asymptomatic fistulae may be treated conservatively with volume reductive drugs such as diuretics. Close monitoring is warranted in this case.[14]

In our case, the fistula's gradient was 130 mmHg, its diameter was ~2 mm, and there was no right ventricular or atrial dilation. The patient was asymptomatic and showed haemodynamic stability throughout the course of his hospitalization. The initial therapeutic decision was to treat conservatively with diuretics and close monitoring. One month later, the patient was still asymptomatic. The echocardiography showed worsening of the fistula (increase diameter, jet, and gradient). Under normal conditions, the gradient should decrease while the diameter of the fistula increases. Increasing of both can only be explained by the excessive amount of blood volume running from the fistula, which creates a higher gradient even in a larger tunnel. The increase in fistula diameter could be explained by the higher accuracy of the CT scan compared with the TTE. While the fistula diameter might have been stable, the increase in the gradient and jet could be explained by a state of volume overload. The patient was completely asymptomatic. Due to lack of strong evidence,[14] and the atypical fistula location, the Heart team decision was to closely monitor the patient for symptoms onset and to reassess fistula closure using a percutaneous approach.

Conclusion

Intra-cardiac fistulas are rare and unpredictable complications post-TAVI. Their mechanism is not well understood. Left ventricular outflow tract to RA fistula is a rare complication post-TAVI. Treatment

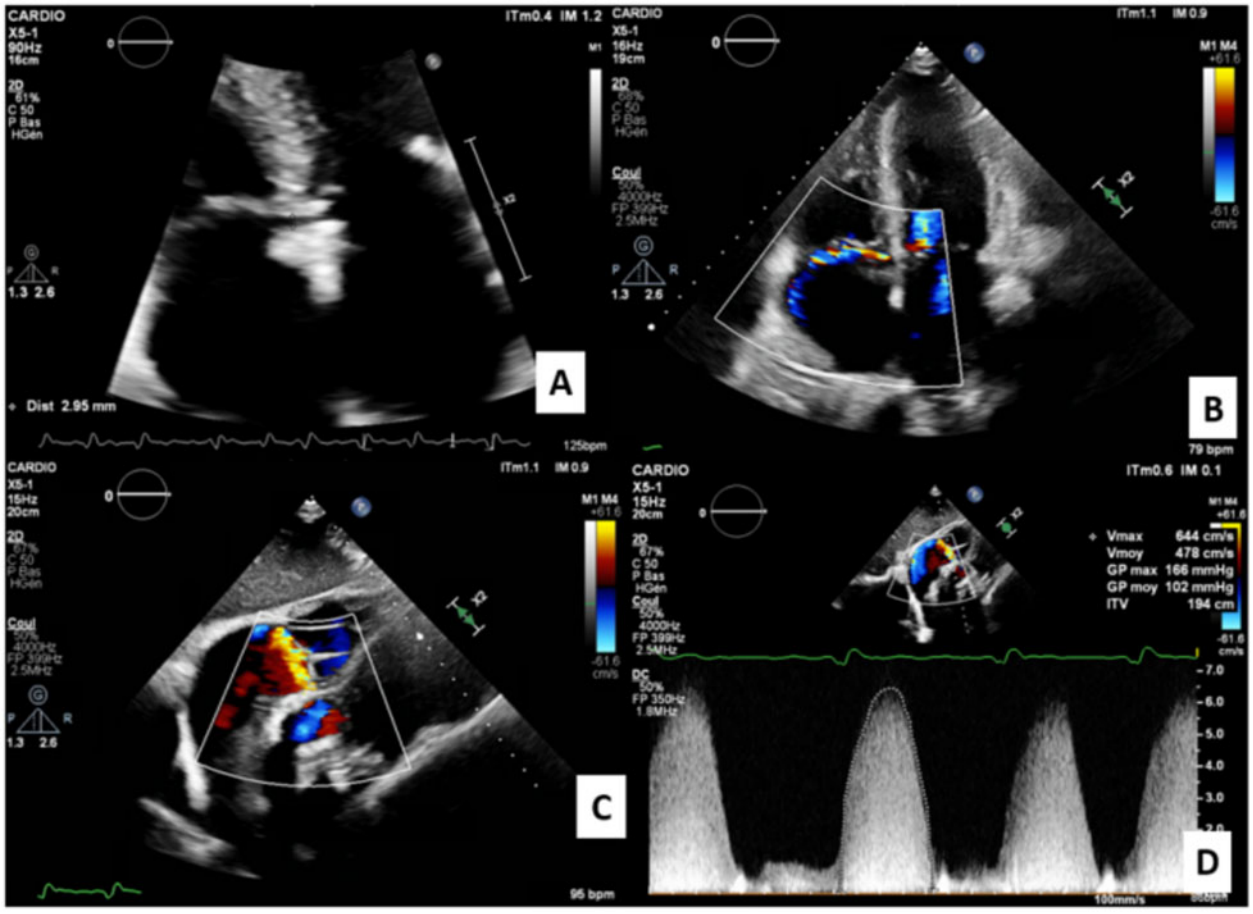
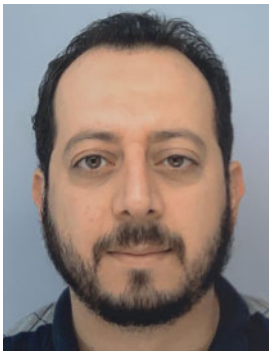


Figure 5 (*A*) Transthoracic echocardiography (zoomed apical four-chamber view) showing the aorto-atrial fistula measuring 2.95 mm). (*B* and *C*) Apical four-chamber and subcostal views showing the left ventricular outflow tract-atrial fistula jet lining the plane of the tricuspid valve with a Coanda effect over the right atrial wall. (*C*) Continuous Doppler showing fistula maximal gradient of 166 mmHg.

options vary from watchful waiting to percutaneous or surgical closure. Alcohol septal ablation before TAVI for patients with ASH >15 mm and dynamic obstruction associated with AS has been proposed as a new treatment strategy for such patients.

Lead author biography



Dr Ahmad Al Ayouby is an interventional cardiologist. He received his general cardiology diploma from the Lebanese University in Beirut in 2017. He started his interventional and structural cardiology formation in Brest University Hospital starting from May 2018 till present day during which he received Interuniversity Diplomas in interventional (University Paris Descartes, 2019) and structural cardiology (University of Lille, 2020).

Supplementary material

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

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

Consent: The authors confirm that written consent for submission and publication of this case report, including images and associated text, has been obtained from the patient in line with COPE guidance.

Conflict of interest: Prof. M.G. receives research fees from Medtronic and Edwards Lifesciences. All other authors have no competing interests.

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