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CASE REPORT
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Case Report: Coil Occlusion of Two Congenital Coronary Cameral Fistulas Connecting Right and Left Circumflex Arteries to the Right Ventricle: An Innovative Stent-Assisted Technique

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Background: Coronary cameral fistulas (CCFs) are rare congenital malformations consisting of abnormal vascular connections between coronary arteries and cardiac chambers, often incidentally found during cardiac catheterizations.

Case summary: A 66-year-old female asymptomatic patient, without cardiovascular risk factors and a history of varicose veins lower extremities and coronavirus disease 2019 (COVID-19) pneumonia in December 2020, was diagnosed by coronary angiography with two large coronary cameral fistulas connecting the distal right coronary artery (RCA) and the distal left circumflex artery (LCx) to the right ventricle (RV). Additional imaging modalities such as two-dimensional transthoracic/transesophageal echocardiography and three-dimensional multidetector CT angiography were required to confirm the fistula's pathway (location, number, and size), which was difficult to delineate using selective coronary angiography alone. After heart team discussion, with the aim to reduce the risk of embolization, an innovative stent-assisted coil occlusion antegrade technique was used with optimal immediate results.

Discussion: Even though our otherwise asymptomatic patient was not the best suitable candidate for an interventional procedure (large vessels, multiple fistulas without distal narrowing, distal portion of the fistula not accessible with the closure device), the innovative stent-assisted fistula coil occlusion technique to stabilize the first coil and deploy safely the additional ones resulted to be key for successful and complete obliteration of the abnormal congenital vascular connections.

Keywords: coronary cameral fistulas, coronary fistulas, coronary angiography, congenital heart disease, coil occlusion

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INTRODUCTION

Coronary cameral fistulas (CCFs) are abnormal vascular connections between coronary arteries and cardiac chambers, reported to be found in ∼0.09–0.5% of unselected patients undergoing diagnostic coronary angiography (1). These fistulas are rare clinical entities, mostly congenital or acquired following trauma or invasive cardiac procedures (endomyocardial biopsy, pacemaker implantation, cardiac surgery). Small fistulas are usually silent and are discovered incidentally on angiography, while large fistulas can present with a continuous murmur and are diagnosed secondary to complications including myocardial infarction and heart failure (2). Usually, coronary fistulas are closed with selected occluders according to their morphology from the venous side through an arteriovenous loop (3, 4). Here, we describe a case of a female patient, who was diagnosed with two large CCFs connecting the distal right coronary artery (RCA) and the distal left circumflex artery (LCx) to the right ventricle (RV) successfully treated by direct arterial approach with a new percutaneous technique.

CASE PRESENTATION

A 66-year-old female patient without cardiovascular risk factors and a history of varicose veins lower extremities and coronavirus disease 2019 (COVID-19) pneumonia in December 2020 underwent a clinical evaluation for an unexplained heart murmur. She was in New York Heart Association class I and her physical examination was negative except for grade II/VI non-radiating continuous murmur, louder in diastole, in the fourth intercostal space, and visible varicose veins in the lower limbs with skin pigmentation. Electrocardiogram showed right bundle branch block and chest X-ray revealed signs of mildly increased pulmonary flow and right chambers dilatation. Two-dimensional (2D) transthoracic/transesophageal Echocardiography (TTE/TEE) color Doppler demonstrated preserved left ventricular ejection fraction, mild mitral regurgitation, RV overload (end-diastolic diameter of 3.2 cm), tricuspid annular plane systolic excursion (TAPSE) of 2.1 cm, slightly enlarged right atrium (dimensions 5.5 × 4.3 cm, area 18 cm^2), significant compression of the right ventricular cavity by a thin-wall round-shaped chamber resulting in dislocation of the tricuspid valve structures toward the ventricular septum with marked tricuspid flow reduction (**Figures 1A–C**). The RV apical four-chamber view showed two systolic-diastolic flows with a width of 7 and 5 mm, respectively, with a pressure gradient of 46 mm Hg and moderate tricuspid regurgitation and an estimated pulmonary pressure of 40 mmHg. The anterior cusp of the tricuspid valve was lengthened and attached closer to the outlet tract, while the septal cusp was moderately displaced, the width of the transtricuspid flow was 1.4 cm, the intraventricular pressure gradient was 16 mm Hg.

Three-dimensional multidetector CT angiography (3D-MDCTA) confirmed the presence of severely dilated and tortuous distal RCA and LCx (**Supplementary Video 1**) and precisely defined entry point of the distal right coronary fistula and two drainage sites draining into the a round-shaped chamber within the right ventricle (**Figures 1D,E**, **Figure 2**).

The patient underwent cardiac catheterization and selective coronary artery angiography in multiple projections showing severely dilated and tortuous RCA and LCx draining distally in a large round-shaped chamber within the RV (**Figure 3**, **Supplementary Videos 2, 3**) confirming the diagnosis of two CCFs connecting to the RV. After the heart team discussion, the decision to proceed with a catheter-based treatment was confirmed to solve the tricuspid flow compression and prevent fistula-related complications, particularly rupture or endoarteritis. Written informed consent, after explanation, was obtained from the patient. Under mild sedation, 2D TEE color Doppler guidance, and local anesthesia, both radial arteries and the right femoral artery were cannulated. We elected to close the large fistulas *via* a direct arterial approach with a new stent-assisted coil occlusion technique to avoid the risk of embolization. The RCA was cannulated using a 6-Fr Judkins Right 4 guiding catheter and a 0.014 in Fielder guidewire (Abbott Vascular) was advanced distally and over it, a Medtronic Resolute Onyx™ DES 5 × 15 mm (Medtronic)was placed in the target point (third segment of RCA); concomitantly, a Cook Mreye (*r*) Flipper coil 40 × 5 mm through 6-Fr multipurpose catheter with stylet inside was placed distally to Onyx™ stent not taking out the stylet from the Flipper to prevent coiling. Then, the balloon of the stent was inflated and the stylet was pulled out from the Mreye (*r*) Flipper system, getting the system more stable and preventing the stent deformation. The Flipper coil was fixed to the arterial wall, the balloon of the stent deflated and taken out and finally, the coil was pushed ahead and detached. Due to the persistent coronary blood flow across the first implanted coil, two additional 8 cm × 5 mm Gianturco coils were deployed proximally to the Flipper coil achieving complete occlusion of the fistula without compromising the flow in coronary side branches (**Figures 4A–D**, **Supplementary Figure 2**). Likewise, the left coronary artery was cannulated using a 5-Fr through a Cordis extra back-up (XB) 3.5 guiding catheter (Cordis) and a Resolute Onyx™ DES 4.0 × 18 mm was implanted in the target point (mid-segment of LCx) pinning down the Cook Mreye ® Flipper coil delivered through 5-Fr Cobra C1 with stylet inside. Subsequently, keeping the 0.014 in Fielder guidewire in the distal segment and withdrawing the balloon of the stent, Flipper's stylet was withdrawn and the Flipper coil pushed distally near the stent. Two additional Gianturco coils (10 cm × 5 mm + 8 cm × 5 mm) were deployed proximally to the Flipper coil. Postprocedure left coronary angiogram showed abolition of the coronary flow by the implanted coils (**Figures 4E–H**, **Supplementary Figure 2**).

The patient was discharged 2 days after the procedure in good clinical conditions. At 3-month follow-up, 2D TTE color Doppler showed decompression of the round-shaped chamber, marked increase in the transtricuspid flow due to the tricuspid valve structures re-expansion with significant improvement of right ventricular systolic function (**Supplementary Figure 1**).

DISCUSSION

The persistence of embryonic sinusoids that perfuse primitive myocardium may lead to a fistulous connection between

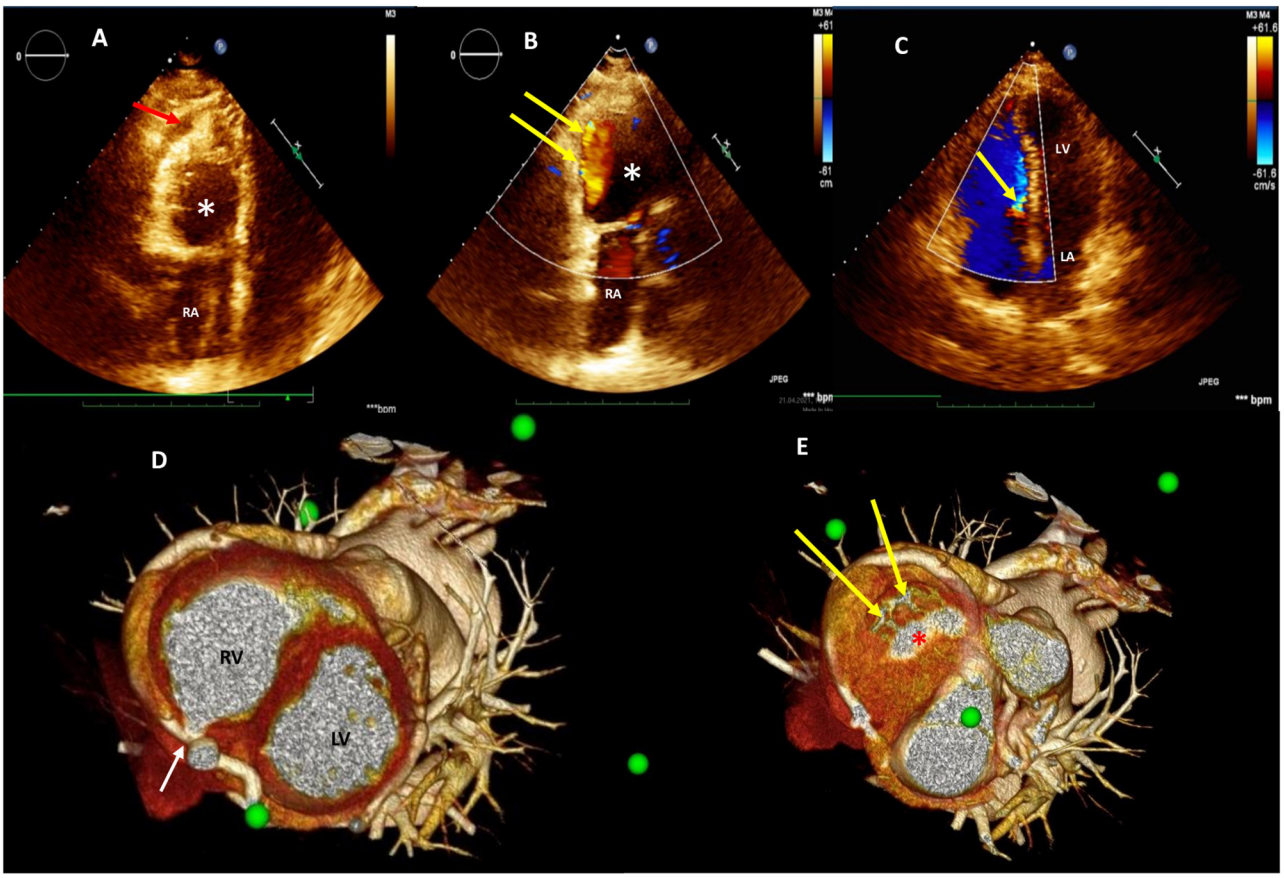

FIGURE 1 | Two-dimensional (2D) transthoracic echocardiogram (TTE) color Doppler showing significant deformation and compression of the right ventricular cavity (red arrow) due to the presence of additional thin-wall round-shaped chamber (white asterisk) **(A)**; two coronary-right ventricle fistulas draining into the additional round-shaped chamber **(B)**; dislocation of the tricuspid valve structures toward the ventricular septum with marked tricuspid flow reduction (yellow arrow) **(C)**; pre-operative assessment by three-dimensional multidetector CT angiography (3D-MDCTA) volume-rendering reconstruction showing entry point (white arrow) **(D)** and two exit sites (yellow arrows) draining into a round-shaped chamber (red asterisk) **(E)** within the right ventricle. 3D-MDCTA, three-dimensional multidetector computed tomography angiography; RA, right atrium; RV, right ventricle; LA, left atrium; LV, left ventricle.

the coronary arteries and cardiac chambers (5). CCFs have been described as arterio-luminal, where there is direct communication with the cardiac chamber concerned, or arterio-sinusoidal, where arterial blood communicates with the cardiac chambers via a sinusoidal network. The majority of CCFs communicate with the right-sided chambers of the heart (55%) and in the remainder of cases with the left side of the heart (35%) or with both (5%) (6–8). Fistulas usually arise predominantly from one of the two major coronary arteries, however, in a small proportion of cases (5%) communications may arise from both coronary arteries like in our case (1, 6).

Coronary cameral fistulas (CCFs) tend to be particularly torturous, presenting a challenge for antegrade transcatheter closure due to the rigidity of the delivery system through the tortuous coronary arteries. Furthermore, there may be multiple feeding arteries to a single drainage point and multiple exit sites may exist, like in this case.

Obliteration by epicardial and endocardial ligations has been the standard surgical treatment (9), generally reserved for large symptomatic fistulas associated with angina or heart failure, multiple communications, very tortuous pathways, and multiple terminations. In addition to infections, arrhythmias, and bleeding, postoperative complications may include myocardial ischemia in the case of coronary ligation.

Suitable candidates for interventional procedures are patients with proximal, easily accessible non-tortuous fistula origin provided there is a single narrow draining site that can be safely accessible. Various methods for closure have been used such as coils, detachable balloons, duct occluders, vascular plugs, covered stents, polyvinyl alcohol foam, and histoacryl resin (10–15).

Our case is unusual for many reasons. Firstly, the asymptomatic patient presented without cardiac or valvular anomalies other than the CCFs. Secondly, multiple imaging modalities were required to confirm the fistula's pathway, which was difficult to delineate using selective coronary angiography alone. Thirdly, the anatomy of those CCFs (tortuous and large vessel, multiple fistulas without distal narrowing, distal portion of the fistula not accessible with the closure device)

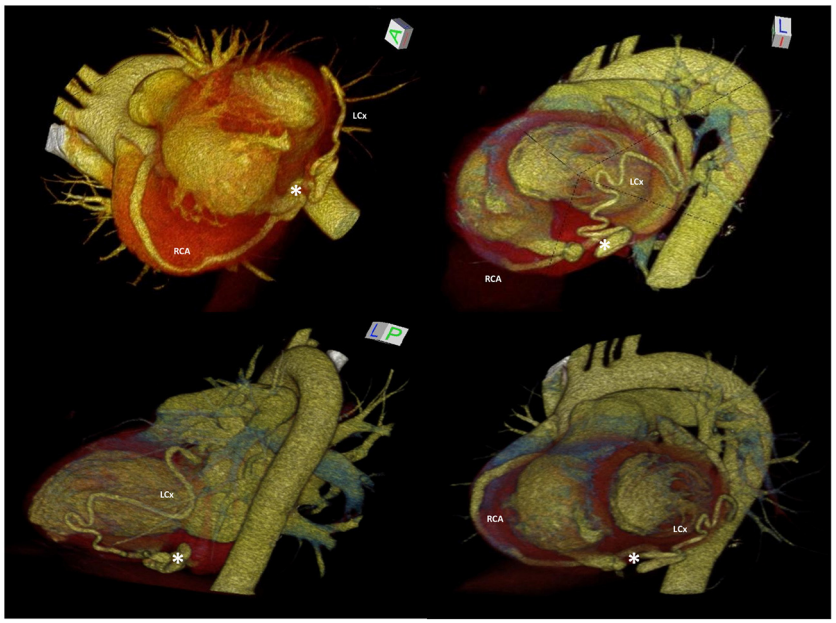

FIGURE 2 | 3D-MDCTA volume-rendering reconstruction showing dilated and tortuous right coronary artery and left circumflex artery forming at the base of the heart a distal anastomosis (white asterisk), followed by drainage into the right ventricle through the posterior-lateral wall. 3D-MDCTA, three-dimensional multidetector computed tomography angiography; LM, left main coronary artery; RCA, right coronary artery; LAD, left anterior descending; LCx, left circumflex.

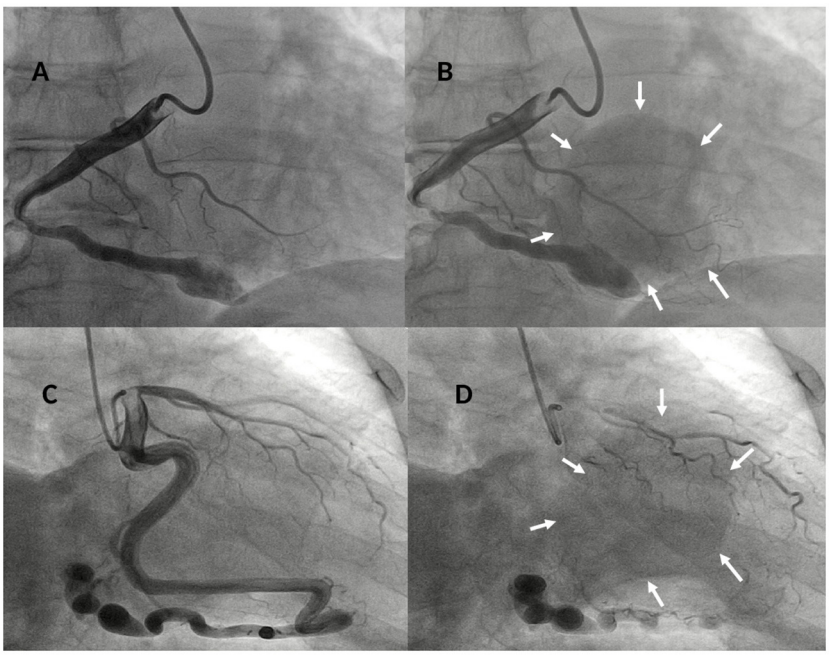

FIGURE 3 | Baseline right coronary angiogram in LAO 30°- caudal 8° view showing a dilated and tortuous coronary artery **(A)** draining in a large round-shaped chamber (yellow arrows) within the right ventricle **(B)** confirming the diagnosis of a right coronary cameral fistula. Baseline left coronary angiogram in RAO 15°- caudal 25° view showing a giant and very tortuous left circumflex (LCx) coronary artery **(C)** draining through a fistulous path in a large round-shaped chamber (yellow arrows) within the right ventricle **(D)** confirming the diagnosis of a left circumflex coronary cameral fistula.

was not suitable for transcatheter closure making coil occlusion at very high risk of embolization. Furthermore, this case of stent-assisted coil occlusion technique to stabilize the first coil was key for success and might broaden the scope of management of large CCFs, which were previously not suitable for device closure.

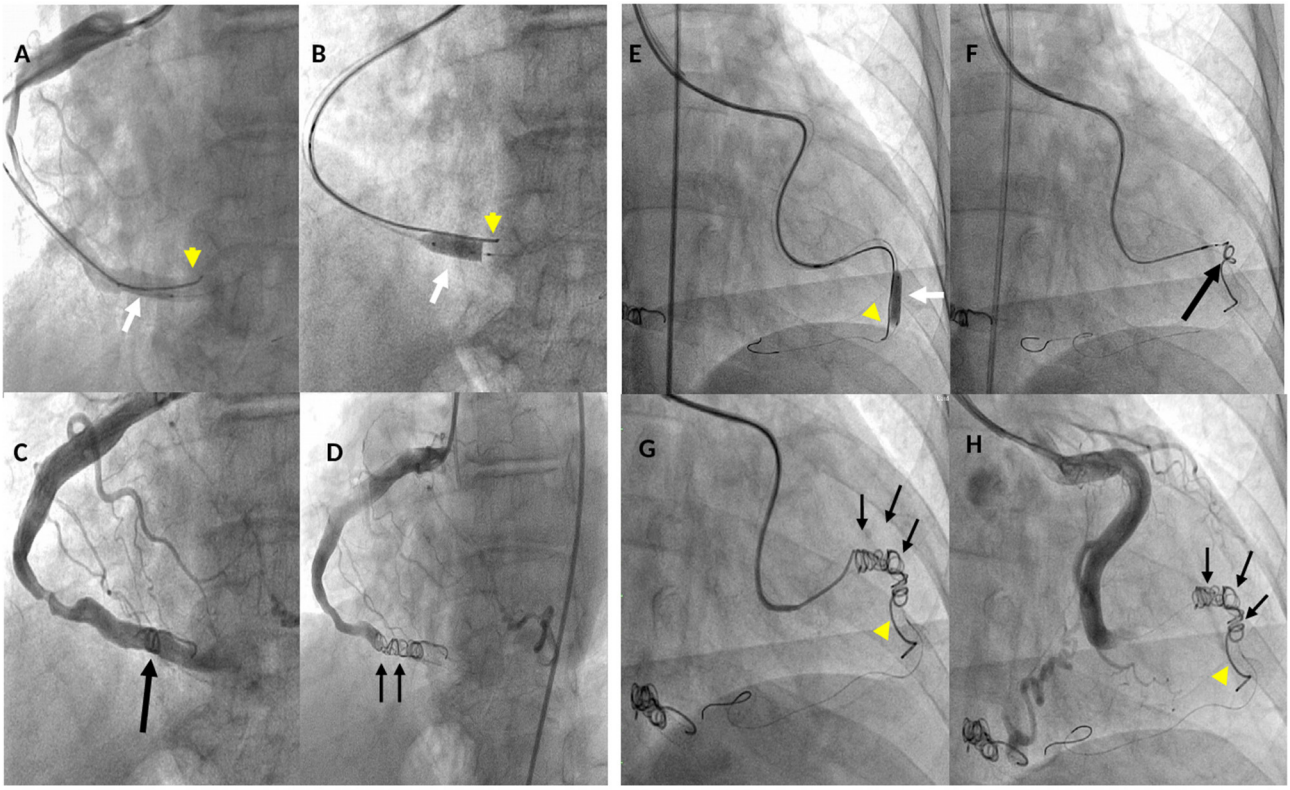

FIGURE 4 | Fluoro-angiographic procedural steps of the coil occlusion of the right coronary (LAO 30° - caud 8°) and left circumflex (RAO 6° - caud 1°) coronary cameral fistulas. **(A)** through a 6-Fr guiding catheter, a Medtronic Resolute Onyx™ DES 5 × 15 mm (white arrow) and the Cook Mreye(*r*) Flipper coil 40 × 5 mm through 6-Fr multipurpose catheter with stylet inside (yellow arrowhead) are concomitantly in place in the distal segment of the right coronary artery (RCA); **(B)** Resolute Onyx™ DES opened and the Cook Flipper coil pinned down by the stent (yellow arrowhead); **(C)** the Flipper coil is detached and pushed ahead to the stent (black arrow) but still there is residual coronary blood flow; **(D)** two additional 8 cm × 5 mm Gianturco coils (black small arrows) deployed proximally to the Flipper coil achieving complete occlusion of the fistula; **(E)** through a Cordis extra back-up (XB) 3.5 guiding catheter, Resolute Onyx™ DES 4.0 × 18 mm was opened (white arrow) pinning down the Cook Mreye(*r*) Flipper coil (yellow arrowhead) delivered through 5-Fr Cobra C1 with stylet inside; **(F)** balloon of stent withdrawn, 0.014 inch coronary wire in the distal LCx, Flipper's stylet withdrawn and the Flipper coil pushed distally near the stent; **(G)** two additional Gianturco coils (10 cm × 5 mm + 8 cm × 5 mm) deployed proximally to the Flipper with effective closure; **(H)** final left coronary angiogram showing abolition of the coronary flow in the distal segment of the LCx by the implanted coils. LAO, left anterior oblique; RAO, right anterior oblique.

CONCLUSION

Coronary cameral fistulas (CCFs) are rare congenital clinical entities, often incidentally found during coronary angiography. The diagnosis can be challenging requiring multiple imaging modalities. The innovative transcatheter stent-assisted coil occlusion technique has been key for successful antegrade closure of those uncommon coronary cameral fistulas, avoiding the risk of coils migration within the coronary artery branches.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/**Supplementary Material**, further inquiries can be directed to the corresponding author.

ETHICS STATEMENT

Written informed consent was obtained from the individual for the publication of any potentially identifiable images or data included in this article.

AUTHOR'S NOTE

This article was the original work of the authors who have all seen and approved of the paper and authorship. The article has not been published elsewhere and is not under consideration in any other journals.

AUTHOR CONTRIBUTIONS

All authors listed have made a substantial, direct, and intellectual contribution to the work and approved it for publication.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fcvm.2021.769235/full#supplementary-material

Supplementary Video 1 | Three-dimensional multidetector computed tomography angiography (3D-MDCTA) showing the presence of severely dilated and tortuous distal RCA and LCx forming at the base of the heart a single anastomosis.

Supplementary Video 2 | Baseline selective right coronary artery angiogram (LAO 30°- caudal 8° view) showing a dilated and tortuous coronary artery draining in a large round-shaped chamber within the right ventricle, confirming the diagnosis of a congenital right coronary cameral fistula.

Supplementary Video 3 | Baseline selective left coronary artery angiogram (RAO 15°- caudal 25° view) showing a tortuous distal left circumflex coronary artery draining through a fistulous path in a large round-shaped chamber within the right ventricle, confirming the diagnosis of a congenital left circumflex coronary cameral fistula.

Supplementary Figure 1 | Two-dimensional (2D) Transthoracic Echocardiogram (TTE) color Doppler in apical four-chamber view showing decompression of the round-shaped chamber (white asterisk, small yellow arrows) **(A)** and marked increase in transtricuspid flow (yellow arrow) **(B)** due to the tricuspid valve structures re-expansion. RA, right atrium; LA, left atrium; RV, right ventricle; LV, left ventricle.

Supplementary Figure 2 | Selective right coronary artery (LAO 30°- caudal 8° view) **(A)** and left coronary artery angiograms (RAO 15°- caudal 25° view) **(B)** showing abolition of the coronary flow (white arrows) in the distal segments of the RCA and the LCx by the implanted coils (orange arrows).

REFERENCES

1. Yuksel S, Yasar E, Nar G, Gulel O, Demircan S, Yilmaz O, et al. Prevalence and characteristics of coronary-cameral communications in adult patients: coronary angiographic analysis of 16,573 patients. *Med Princ Pract.* (2014) 23:336–9. doi: 10.1159/000363183
2. Qarajeh R, Qureini A, Thompson RC, Saeed IM. Acute heart failure from coronary cameral fistula. diagnostic challenge and therapeutic opportunity. *J Am Coll Cardiol Case Rep.* (2020) 2:2368–71. doi: 10.1016/j.jaccas.2020.09.034
3. Carminati M, Giugno L, Chessa M, Butera G, Piazza L, Bussadori C. Coronary-cameral fistulas: indications and methods for closure. *EuroIntervention.* (2016) 12:X28–X30. doi: 10.4244/EIJV12SXA6
4. Mokgwathi GT, Ogunbanjo GA. The management of a coronary cameral fistula: lessons learnt. *South Afr J Anaesth Analg.* (2012) 18:168–70. doi: 10.1080/22201173.2012.10872847
5. Challoum D, Pericleous A, Dimitrakaki IA, Danelatos C, Dimitrakakis G. Coronary arteriovenous fistulae: a review. *Int J Angiol.* (2014) 23:1–10. doi: 10.1055/s-0033-1349162
6. Boyle S, Jesuthasan LSB, Jenkins C, Challa P, Ranjan S, Dahiya A. Coronary-cameral fistula. *Circ Cardiovasc Imaging.* (2019) 12:8691. doi: 10.1161/CIRCIMAGING.118.008691
7. Khan Minhas A, Haq E, Awan AA, Khan AA, Qureshi G, Balakrishna P. Coronary-cameral fistula connecting the left anterior descending artery and the first obtuse marginal artery to the left ventricle: a rare finding. *Case Rep Cardiol.* (2017) 2017:8071281. doi: 10.1155/2017/8071281
8. Maknojia A, Pride Y, Ghatak A, Lee J. Fistula between the first obtuse marginal branch of the left circumflex and the left ventricular cavity: a rare anomaly. *Cureus.* (2021) 13:e13316. doi: 10.7759/cureus.13316
9. Kamiya H, Yasuda T, Nagamine H, Sakakibara N, Nishida S, Kawasuji M, et al. Surgical treatment of congenital coronary artery fistulas: 27 years' experience and a review of the literature. *J Card Surg.* (2002) 17:173–7. doi: 10.1111/j.1540-8191.2002.tb01195.x
10. Krabill KA, Hunter DW. Transcatheter closure of congenital coronary arterial fistula with a detachable balloon. *Pediatr Cardiol.* (1993) 14:176–8. doi: 10.1007/BF00795650
11. Fischer G, Apostolopoulou SC, Rammos S, Kiaffas M, Kramer HH. Transcatheter closure of coronary arterial fistulas using the new Amplatzer vascular plug. *Cardiol Young.* (2007) 17:283–7. doi: 10.1017/S1047951107000510
12. Mullasari AS, Umesan CV, Kumar KJ. Transcatheter closure of coronary artery to pulmonary artery fistula using covered stents. *Heart.* (2002) 87:60. doi: 10.1136/heart.87.1.60-a
13. Nkya DA, Sinyangwe G, Takawira FF. Transcatheter occlusion of giant congenital coronary cameral fistulae: a case series. *J Med Case Rep.* (2019) 13:307. doi: 10.1186/s13256-019-2254-x
14. Bruckheimer E, Harris M, Kornowski R, Dagan T, Birk E. Transcatheter closure of large congenital coronary cameral fistulae with Amplatzer devices. *Catheter Cardiovasc Interv.* (2010) 75:850–4. doi: 10.1002/ccd.22365
15. Karagoz T, Celiker A, Cil B, Çekirge S. Transcatheter embolization of a coronary fistula originating from the left anterior descending artery by using N-butyl 2-cyanoacrylate. *Cardiovasc Intervent Radiol.* (2004) 27:663–5. doi: 10.1007/s00270-004-0251-x

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CASE REPORT
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Transradial Retrograde Percutaneous Coronary Intervention of Chronic Total Occlusion *via* an Ipsilateral Septal Collateral Using a Single Guiding Catheter: A Case Report

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Background: With the development of specialized equipment and the retrograde technique, success rates for percutaneous coronary intervention (PCI) of chronic total occlusions (CTOs) have increased from 60 to 90% in the past 10 years. Performing PCI *via* a collateral channel from the contralateral artery, using two guiding catheters, is usually the preferred approach to retrograde CTO-PCI. In the case described in this report, only the ipsilateral septal collateral artery from the proximal occluded left anterior descending (LAD) artery was available. The procedure can be performed successfully from radial artery access using a single guiding catheter.

Case Summary: A 57-year-old patient, with a history of anterior and inferior myocardial infarction and previous PCI, underwent a planned coronary arteriography due to his complaints of typical angina symptoms. Coronary angiography revealed stent occlusion located mid-LAD and severe in-stent restenosis in the distal right coronary artery (RCA). A proximal septal branch was supplying the distal LAD retrogradely. After repeated failed attempts at antegrade PCI for the LAD's CTO, the retrograde approach was tried. This intervention finally succeeded through the ipsilateral septal collateral. It was performed *via* a single radial-artery access throughout the whole process. Post-operatively, the patient had no complications and was stable at 1-year follow-up.

Conclusion: The transradial approach to retrograde PCI for CTO *via* an ipsilateral septal collateral using a single guiding catheter is feasible and safe in appropriately selected cases.

Keywords: chronic total occlusion, percutaneous coronary intervention, retrograde approach, radial artery access, case report

INTRODUCTION

Successful percutaneous coronary intervention (PCI) of chronic total occlusions (CTOs) has been associated with a reduced need for coronary artery bypass graft surgery (CABG), improvements in left ventricular function, and better long-term survival (1). With the development of specialized equipment and the retrograde technique, success rates for recanalization of CTOs have increased from 60 to 90% in the past 10 years (2, 3). The advantages of the retrograde approach in CTO-PCI

have been observed in many clinical cases. Using septal collaterals from the contralateral artery as an access route shows a high success rate in retrograde CTO-PCI. However, not all CTO cases are well-suited for using the contralateral septal collateral as an access route. In some cases of left anterior descending (LAD) lesions, only the ipsilateral septal collateral from the same artery is observed to supply the distal recipient artery (4).

Femoral arterial access is usually the preferred approach to CTO-PCI because of its ability to firmly support a larger guiding catheter (7-French), but in several recent reports, radial access has been demonstrated to be possible, safe, and effective (5). We present a case of mid-LAD CTO recanalization performed successfully *via* an ipsilateral septal collateral, using a single guiding catheter that was positioned in the radial artery.

CASE PRESENTATION

A 57-year-old patient was admitted to the cardiovascular department with progressive angina and dyspnea over 6 months. The patient's past medical history included anterior and inferior myocardial infarction, which was treated with PCI of the anterior descending branch and right coronary artery (RCA) 17 years prior, and hypertension. He was on several medications for 3 months, including clopidogrel, aspirin, atorvastatin, metoprolol, telmisartan, furosemide, and spironolactone. The physical examination was notable for rales at the lung bases and mild lower-extremity edema. The remainder of the physical examination was unremarkable.

The electrocardiogram showed pathologic Q-waves in the anterior and inferior leads. Transthoracic echocardiography showed impaired left ventricular function (EF = 38%) and a dilated left ventricle (LV = 75 mm). The patient was diagnosed with coronary heart disease with unstable angina and heart failure. After receiving intensive anti-anginal and anti-heart failure medications, the patient underwent planned coronary arteriography a week later. Coronary angiography showed stent occlusion in the mid-LAD (**Figure 1A**) and severe in-stent restenosis in the distal RCA (**Figure 1B**). A proximal septal branch was supplying the distal LAD retrogradely (**Figure 1A**). The SYNTAX (Synergy between PCI with Taxus and Cardiac Surgery) score was 14.5.

Coronary artery bypass graft (CABG) surgery was advised for the patient according to the coronary angiography. The patient was thoroughly informed of the risks of the CTO-PCI; he refused CABG surgery and accepted the risks of PCI. PCI for the LAD CTO was then attempted. After repeated attempts at antegrade access failed (**Figure 2A**), the retrograde approach was tried. The 6-Fr JL 3.5 guiding catheter of radial artery continued to be used because the patient's radial spasm made changing sheaths difficult. In addition, there were concerns that his poor left-ventricular function would make lying flat over a long period risky; therefore, femoral access was the back-up site if transradial access failed. The retrograde wire (SION black 190, Asahi) was advanced through the septal collateral with the microcatheter (Corsair 150, Asahi), reaching the distal CTO lesion (**Figure 2B**). Then, the retrograde Sion wire was exchanged for a PILOT150 (Abbott) wire. It crossed the CTO segment successfully and

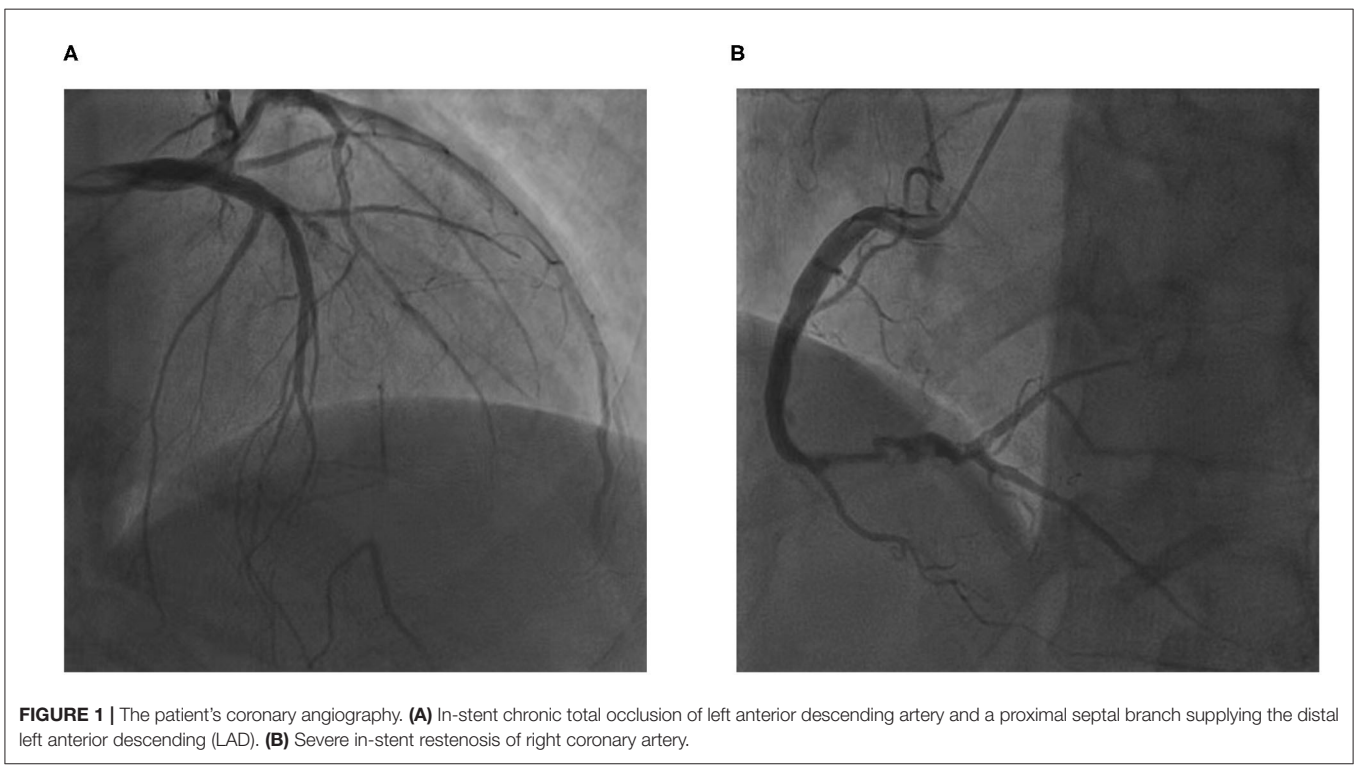

FIGURE 1 | The patient's coronary angiography. **(A)** In-stent chronic total occlusion of left anterior descending artery and a proximal septal branch supplying the distal left anterior descending (LAD). **(B)** Severe in-stent restenosis of right coronary artery.

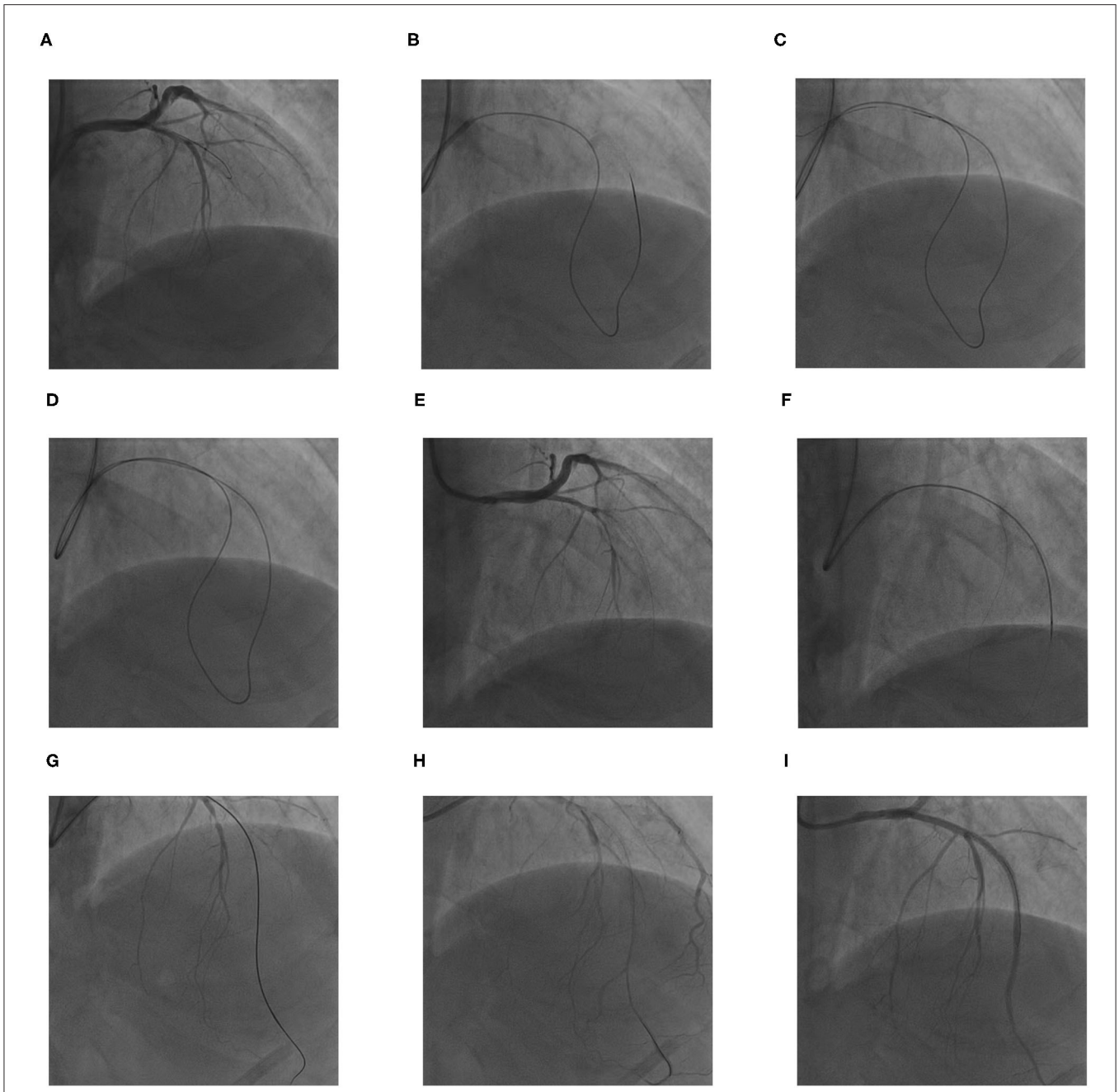

FIGURE 2 | The procedures for retrograde percutaneous coronary intervention (PCI) of LAD chronic total occlusion (CTO). **(A)** The antegrade wire could not cross the CTO lesion. **(B)** The retrograde soft wire was advanced through the septal collateral with the microcatheter reaching the distal CTO lesion. **(C,D)** The retrograde soft wire was exchanged for a stiff wire *via* microcatheter, which crossed the CTO segment into proximal LAD true lumen, and threaded into the antegrade catheter with microcatheter retrogradely. **(E)** A 300-cm RG3 wire was externalized *via* the microcatheter. **(F)** The microcatheter was inserted into the proximal LAD through the septal collateral antegradely; the RG3 wire was removed. **(G,H)** The microcatheter was introduced to the distal LAD; the antegrade wire was advanced through the lesion. When the wire was confirmed in true lumen by angiogram, the microcatheter was removed. **(I)** Three drug-eluting stents were deployed at mid LAD successively.

was threaded into the guiding catheter retrogradely. The microcatheter was subsequently advanced over the wire into the guiding catheter (**Figures 2C,D**). A 300-cm RG3 wire (Asahi) was externalized *via* the microcatheter (**Figure 2E**). Then, the microcatheter was withdrawn retrogradely and advanced to the proximal LAD through the septal collateral over the wire antegradely. The RG3 wire was removed (**Figure 2F**). Then, the microcatheter was introduced to the distal LAD. Afterward, the antegrade wire (SION black) was advanced through the lesion. When the wire was confirmed to be in the true lumen by an angiogram, the microcatheter was removed (**Figures 2G,H**), and three drug-eluting stents were deployed successively at mid-LAD

TABLE 1 | Timeline.

Timeline	Events	Coronary angiography	Intervention
February 2002	Acute anterior and inferior myocardial infarction	1. Acute occlusion of left anterior descending artery (LAD) 2. Severe restenosis of right coronary artery (RCA)	Urgent percutaneous coronary intervention (PCI) of LAD and selective PCI of RCA
November 2019	Typical angina symptoms	1. In-stent chronic total occlusion (CTO) of LAD 2. Severe in-stent restenosis of RCA	Retrograde CTO PCI of LAD and selective PCI of RCA
December 2020	1-year follow-up: symptom free	Absence	Drug therapy

(**Figure 2I**). The final angiogram showed Thrombolysis in Myocardial Infarction (TIMI) flow grade 3 in the distal LAD.

After the intervention, the patient continued to be treated with guideline-directed medical therapy. The patient had no complications and underwent selective PCI of the RCA 2 weeks later. The patient was stable over 1 year of follow-up appointments. A detailed timeline of the events and therapy of the patient is provided (**Table 1**).

DISCUSSION

Chronic total occlusion (CTO) is defined as an occlusive lesion of over 3-month duration. The CTO-PCI tends to be a challenge for surgeons because of its low success rate and lengthy procedure. The main cause of CTO-PCI failure is usually an inability to cross the lesion (6). Improvements in the retrograde approach have helped increase the success rate. The proximal fibrous cap of a CTO lesion is characteristically hard and thick (7), which may increase the failure rate and the risk of subintimal dissection, while the distal cap is thinner and tapered so that the wire can cross it easily. This provides a theoretical basis for retrograde PCI. In the case described here, a large septal branch arising from the proximal LAD CTO was visible; otherwise, mid-LAD had a long occlusion. These observations suggested that retrograde PCI would be relatively easier to accomplish when antegrade attempts failed repeatedly.

Selecting a suitable collateral channel is the key to the success of retrograde approach. The selectable collateral channel for retrograde PCI could be a septal collateral or an epicardial collateral. Septal collaterals are preferred in majority of cases because, compared to the epicardial collaterals, they usually have a shorter and less tortuous course. Furthermore, they are less likely to cause tamponade when they are injured (8). In this case, no septal collateral from the RCA to the LAD was observed due to the severe stenosis in RCA. Instead, a septal collateral from the proximal LAD was found filling the distal LAD retrogradely, so it was chosen to be the retrograde access.

In most retrograde PCI cases, surgeons use the double-guiding catheter technique (9), but a single-guiding catheter was used *via* the radial artery in this case because of the patient's radial spasm and his poor left ventricular function. This reduced his time lying flat and minimized the risk of the onset of acute heart failure. The transradial approach could lower the risk of access-site complications, which include femoral artery hematoma and bleeding, and the risk of the onset of acute heart failure for patients with impaired heart function. Additionally, it may increase patient comfort and shorten hospital stays.

Nevertheless, this approach has limitations. First, it is the fact that smaller-size guiding catheters in the radial artery are less supportive and harder to operate. Second, when multiple devices need to be used simultaneously, a larger guiding catheter is more appropriate. Therefore, radial CTO PCI with a single-guiding catheter can be effective in appropriately selected cases. Third, if the retrograde wire has difficulty crossing the CTO segment or the microcatheter is difficult to advance across the CTO segment, the controlled antegrade and retrograde tracking (CART) technique, Knuckle technique, or anchor-balloon technique would be needed. In such cases, a second arterial access and a large-size guiding catheter would be required. Fourth, compared to the RG3 wire externalization, the "rendez-vous" technique may be more efficient and safer, but it is not straightforward to perform this operation with a single, small-caliber catheter like the 6-Fr guiding catheter. Therefore, the RG3 wire externalization technique was adopted in this case. When the RG3 wire was externalized, the safest method was to pull back the microcatheter up to the distal part of the CTO but keep covering the septal channel with the microcatheter. A new microcatheter was used antegradely and advanced beyond the CTO segment. However, because the 6-FrJL3.5 guiding catheter that was used could not accommodate two microcatheters at the same time, the microcatheter was withdrawn and the same one was used to advance antegradely. Indeed, this procedure risked damaging the uncovered septal collateral channel. To protect the septal collateral, the microcatheter was advanced to the proximal LAD through the septal collateral over the wire antegradely when we removed the RG3 wire.

CONCLUSION

This rare case report demonstrated that transradial approach of retrograde PCI for the CTO *via* an ipsilateral septal collateral using a single guiding catheter is feasible and safe in appropriately selected cases.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author/s.

ETHICS STATEMENT

Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

AUTHOR CONTRIBUTIONS

XL wrote the first draft of the manuscript. JZ wrote sections of the manuscript. HZ and PZ performed the PCI, collected cardiological data, and prepared PCI pictures. NF contributed to the case diagnosis, therapy, and decision-making. All authors contributed to manuscript revision, read, and approved the submitted version.

REFERENCES

1. Sianos G, Barlis P, Di Mario C, Papafaklis MI, Büttner J, Galassi AR, et al. European experience with the retrograde approach for the recanalisation of coronary artery chronic total occlusions. A report on behalf of the euro CTO club. *Eurointervention*. (2008) 4:84–92. doi: 10.4244/EIJV4I1A15
2. Kimura M, Katoh O, Tsuchikane E, Nasu K, Kinoshita Y, Ehara M, et al. The efficacy of a bilateral approach for treating lesions with chronic total occlusions the CART (controlled antegrade and retrograde subintimal tracking) registry. *JACC Cardiovasc Interv*. (2009) 2:1135–41. doi: 10.1016/j.jcin.2009.09.008
3. Zhang B, Wong A. The confluent balloon technique for retrograde therapy of chronic total occlusion. *Catheter Cardiovasc Interv*. (2011) 78:60–4. doi: 10.1002/ccd.22857
4. Egred M. Transradial retrograde recanalization of totally occluded left anterior descending artery using a single 7 fr guiding catheter. *J Invasive Cardiol*. (2013) 25:57–60.
5. Brilakis ES, Grantham JA, Thompson CA, DeMartini TJ, Prasad A, Sandhu GS, et al. The retrograde approach to coronary artery chronic total occlusions: a practical approach. *Catheter Cardiovasc Interv*. (2012) 79:3–19. doi: 10.1002/ccd.23004
6. Rathore S, Katoh O, Matsuo H, Terashima M, Tanaka N, Kinoshita Y, et al. Retrograde percutaneous recanalization of chronic total occlusion of the coronary arteries: procedural outcomes and predictors of success in contemporary practice. *Circ Cardiovasc Interv*. (2009) 2:124–32. doi: 10.1161/CIRCINTERVENTIONS.108.838862
7. Chon MK, Kim JS, Chun KJ. Retrograde percutaneous coronary intervention for left anterior descending chronic total occlusion via an ipsilateral intraseptal collateral channel using a single guiding catheter. *J Coll Physicians Surg Pak*. (2016) 26:S4–6.
8. Surmely JF, Katoh O, Tsuchikane E, Nasu K, Suzuki T. Coronary septal collaterals as an access for the retrograde approach in the percutaneous treatment of coronary chronic total occlusions. *Catheter Cardiovasc Interv*. (2007) 69:826–32. doi: 10.1002/ccd.20816
9. Thompson CA, Jayne JE, Robb JF, Friedman BJ, Kaplan AV, Hettleman BD, et al. Retrograde techniques and the impact of operator volume on percutaneous intervention for coronary chronic total occlusions an early U.S. experience. *JACC Cardiovasc Interv*. (2009) 2:834–42. doi: 10.1016/j.jcin.2009.05.022

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CASE REPORT
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Transjugular Approach to Closure of Patent Foramen Ovale Under the Guidance of Fluoroscopy and Transthoracic Echocardiography: A Case Report

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Background: We describe a rare case of patent foramen ovale (PFO) associated stroke in a patient with pulmonary embolism, inferior vena cava thrombosis and undergoing filter implantation who successfully underwent PFO closure using the right internal jugular venous approach.

Case Summary: This is a rare case of a 42-year-old patient who presented with stroke and pulmonary embolism and was diagnosed with a PFO, inferior vena cava thrombosis and underwent filter implantation. The patient suffered from stroke and pulmonary embolism successively; that is, embolic events occurred in both the arterial and venous systems. Transesophageal echocardiography (TEE) showed a PFO with an atrial septal aneurysm (ASA), which we considered a "pathological" PFO. Due to the obstructive nature of the inferior vena cava approach, we successfully performed PFO closure via the right internal jugular venous approach under the guidance of X-ray and transthoracic echocardiography (TTE).

Discussion: The right jugular venous approach provides a simple technical solution for patients who require PFO closure when femoral venous access is unavailable, which can be performed under X-ray and TTE guidance.

Keywords: patent foramen ovale, inferior vena cava thrombosis, closure, transjugular, atrial septal aneurysm

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INTRODUCTION

Transcatheter closure of a patent foramen ovale (PFO) is usually performed using a femoral venous approach. This procedure becomes more difficult when femoral vein access is not feasible due to congenital or acquired reasons. There are only a few reports of PFO closure via the jugular venous approach in this special case, and these procedures were mostly performed under general anesthesia and under the guidance of transesophageal echocardiography (TEE) or intracardiac echocardiography (ICE) (1–3). We report a case of PFO associated stroke in a patient with atrial septal aneurysm (ASA), pulmonary embolism, inferior vena cava thrombosis and filter implantation, under the guidance of local anesthesia, X-ray and transthoracic echocardiography (TTE), the patient was successfully underwent PFO closure using the right internal jugular venous approach.

CASE PRESENTATION

A 42-year-old male with sudden left upper extremity dysfunction was diagnosed with cerebral infarction by MRI and cerebral angiography. Intracerebral hemorrhage occurred after intravenous thrombolysis. One month later, a CT scan of the pulmonary artery was performed because the patient complained of left chest pain, shortness of breath and lower extremity edema, which indicated extensive thrombosis of the bilateral pulmonary arteries. Ultrasound of bilateral lower extremity veins showed left femoral vein, superficial femoral vein, and popliteal vein thrombosis. To prevent massive pulmonary embolism, anticoagulation therapy (rivaroxaban 20 mg/d) and inferior vena cava filter implantation were performed. Antithrombin III, protein C, protein S, tumor markers, antiphospholipid antibodies, autoantibodies were all negative. Because the patient has successively experienced embolism of the arterial system and venous system without risk factors, such as hypertension, diabetes, smoking and so on, paradoxical embolism cannot be excluded. TEE examination revealed a large PFO with an ASA (**Figure 1**). After multidisciplinary diagnosis, transcatheter PFO closure is recommended.

Preoperative TTE examination showed a PFO with an ASA, and contrast transthoracic echocardiography (cTTE) showed substantial right-to-left shunting (RLS) at rest and Valsalva maneuver (**Figure 2**). The CT examination of the lower extremity veins showed implantation of the inferior vena cava filter, inferior vena cava and bilateral external iliac vein thrombosis, and bilateral internal iliac vein thrombosis (**Figure 3**).

Due to the conventional approach of bilateral external iliac veins, internal iliac veins and inferior vena cava all have thrombi, the superior vena cava and jugular venous systems appeared normal; therefore, a right jugular venous approach was planned. A 6 Fr sheath was placed in the right jugular vein using the Seldinger technique. Initial attempts to probe the PFO utilized multiple catheters (6Fr TERUMO Radial TIG catheter, Medtronic LAUNCHER 6Fr AL catheter, Cordis 6Fr JR4 catheter), and probing with a straight 0.035 inch guidewire was unsuccessful. A Medtronic LAUNCHER 6Fr EBU 3.5 mm guiding catheter was then advanced to the right atrium, initially positioned below the atrial septum at the level of the tricuspid valve, and then withdrawn superiorly, allowing it to engage and pass through the PFO into the left atrium and left superior pulmonary vein (**Figures 4A,B**). Due to insufficient support, when the guiding catheter was passed through the PFO, the guiding catheter and guidewire were slid into the inferior vena cava simultaneously (**Figure 4C**). Under the guidance of the guidewire, the EBU3.5 mm guiding catheter was sent to the fossa ovalis again, and a 180 cm 0.025" stainless steel guidewire with coiled floppy tip was exchanged. The guiding catheter passed through the PFO smoothly and the stainless steel guidewire was looped into the left atrium (**Figure 4D**). The position of the guidewire was confirmed by TTE, and there was no pericardial effusion. Withdrawing the guiding catheter, as per the classic

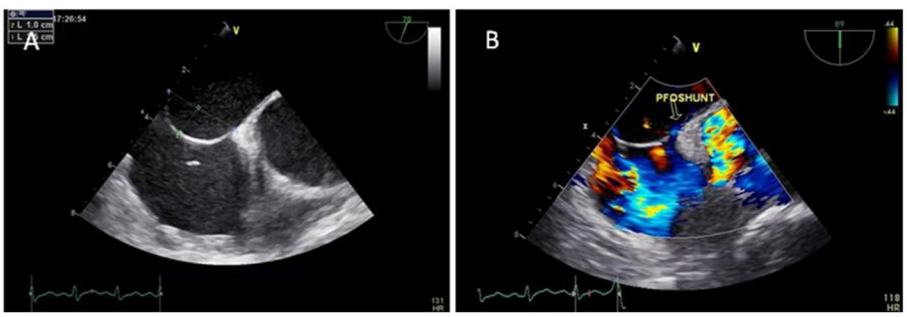

FIGURE 1 | TEE showed a PFO with an ASA **(A)** and a small amount of left-to-right shunt through the fossa ovale **(B)**.

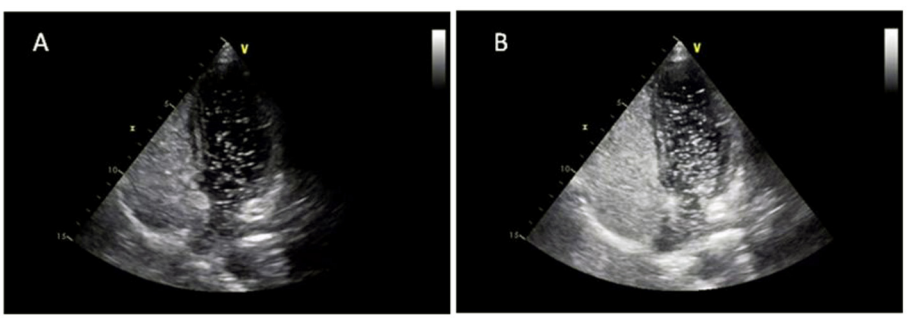

FIGURE 2 | cTTE showed substantial RLS at rest **(A)** and Valsalva maneuver **(B)**.

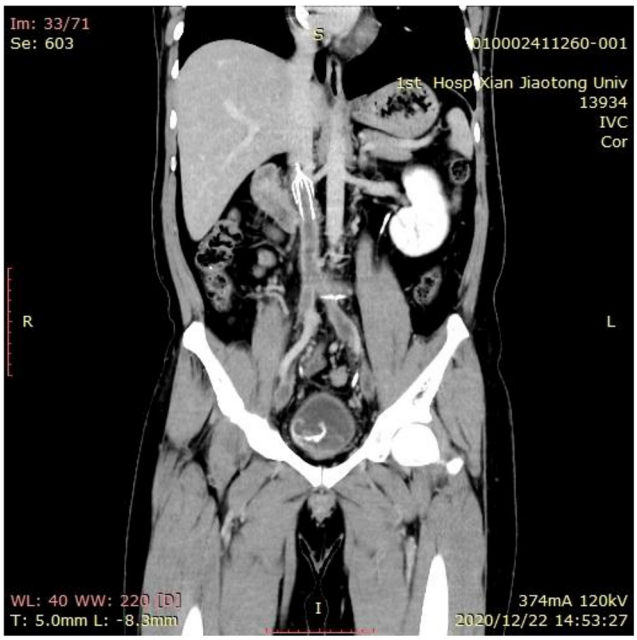

FIGURE 3 | The CT examination of the lower extremity veins showed implantation of the inferior vena cava filter, inferior vena cava and bilateral external iliac vein thrombosis, and bilateral internal iliac vein thrombosis.

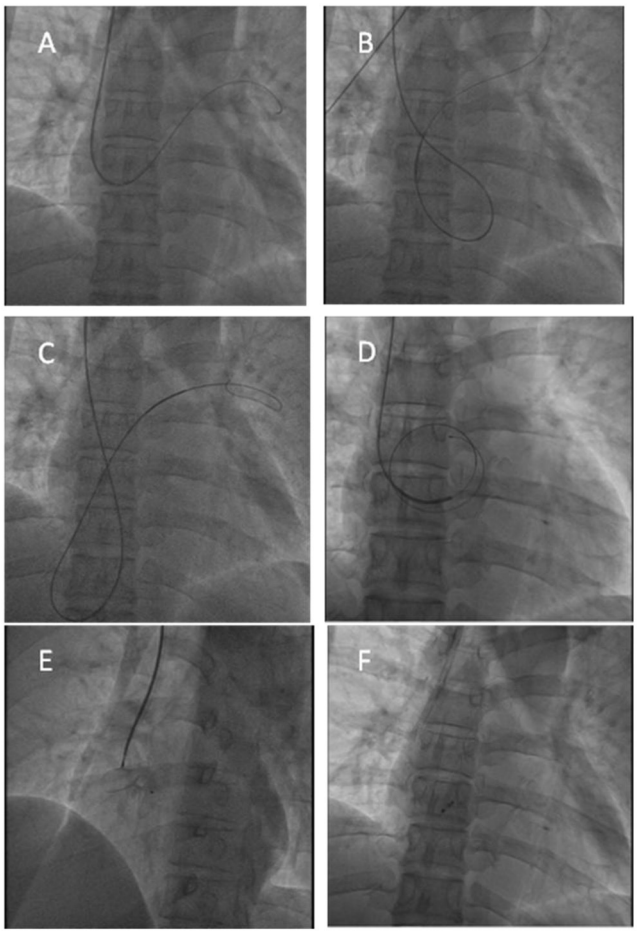

FIGURE 4 | Transcatheter PFO closure procedure. **(A,B)** The guidwire reached the left superior pulmonary vein through the PFO; **(C,D)** The ordinary guidewire could not provide sufficient support, and a 180 cm 0.025" stainless steel guidewire with coiled floppy tip was exchanged to provide sufficient support; **(E)** X-ray showed good position before the occluder was released; **(F)** X-ray showed good position after the occluder was released.

femoral venous approach, a 9Fr Abbott TorqVue 180° delivery sheath was advanced over the wire into the left atrium, and an Amplatzer 30 mm PFO occluder (St. Jude Medical, Golden Valley, MN) was successfully deployed with good apposition as assessed by fluoroscopy and TTE (**Figures 4E,F**). Following device deployment, there was no evidence of residual shunt on TTE Doppler, which also showed a negative bubble study result. Recovery was uncomplicated, and the patient was discharged the next day.

DISCUSSION

Over the past 5 years, multiple randomized controlled trials (RCTs) have established robust evidence for preventing cryptogenic stroke (CS) by percutaneous closure of PFO (4–7). Therefore, the Amplatzer PFO occluder is indicated for percutaneous closure of a PFO to reduce the risk of recurrent ischemic stroke in patients who are predominantly between the ages of 18 and 60 years, who have had CS due to a presumed paradoxical embolism, as determined by a neurologist and cardiologist following an evaluation to exclude known causes of ischemic stroke.

The choice of approach for transcatheter PFO closure usually depends on the anatomical relationship between the vena cava and the atrial septum. Studies have shown that the angle between the inferior vena cava and the PFO tunnel is usually ∼45°, which is very suitable for guiding the guidewire through the PFO to reach the left atrium, thus making it easier for the subsequent delivery sheath system to reach the left atrium, and thereby shortening the procedure time (2). Therefore, the femoral venous approach is often used as a method to easily and reliably pass across the PFO into the left atrium and, subsequently, deploy the device.

When the femoral venous approach is not possible due to inferior vena cava interruption for congenital or acquired reasons, transcatheter PFO closure by conventional methods is difficult to perform. Moreover, the number of PFO closures increases each year. In addition to the femoral venous approach, alternative approaches for PFO closure include the jugular, hepatic, and subclavian venous approaches. PFO closure through these approaches usually needs to be performed under the guidance of general anesthesia and TEE, and there are also individual

reports that it is performed under the guidance of ICE (1–3).

If this patient chose the femoral venous approach, transfemoral-iliac vein intubation and balloon dilation were required first. Due to the increased risk of thrombus in the inferior vena cava along with a high-risk PFO, the risk of paradoxical embolism during the procedure is high, so the femoral venous approach is obviously not feasible. Transhepatic access has been reported in the interventional treatment of atrial septal defect (ASD) and PFO because it does not require special catheter technology (8–10). However, it requires a gelatin sponge or coils to stop bleeding through the ducts that pass through the liver, and there have been individual cases of postoperative abdominal discomfort due to peritoneal irritation caused by bleeding (11). At present, the subclavian venous approach, which has the potential to increase the risk of hemorrhage due to the large-sized sheath placed through the junction of the clavicle and the first rib, is basically not used. For these reasons, we plan to use the internal jugular vein as the preferred access point and the hepatic venous approach as an alternative.

It is technically challenging to approach the fossa ovale after entering the right atrium from the internal jugular vein because the tip of the sheath needs to have a curvature close to 90° or more to be close to the fossa ovale. Existing case reports show that various curved guiding catheters can be tried, the tip of the catheter can be shaped with cold saline, or the flexible sheath can be selected to increase the maneuverability of the catheter with the guidewire to pass through the PFO (1, 2, 12). In addition, the stiffness of the catheter tip also affects the procedure process. If the catheter tip is too soft, even if the guidewire passes through the PFO, the stiffened wire cannot be exchanged, and the delivery sheath cannot pass through the atrial septum due to a lack of strong support. If the tip is too rigid, there is a risk of injuring the left atrial wall when the catheter is passed through the atrial septum into the left atrium.

We describe a PFO associated stroke in a patient with an ASA, pulmonary embolism, inferior vena cava thrombosis and filter implantation who successfully underwent PFO closure by the guidance of X-ray and TTE using the right internal jugular venous approach for the first time. In this patient, we tried different curved catheters and failed to pass through the PFO. Then, we chose the EBU guiding catheter, and with the help of the guidewire, we reached the flaccid atrial septum and entered the left atrium through the PFO. At the same time, to solve the problem of insufficient support, we used a 180 cm 0.025" stainless steel guidewire with coiled floppy tip to pass through the interatrial septum to reach the left atrium and then successfully released the PFO occluder under the guidance of X-ray and TTE.

CONCLUSION

The right jugular venous approach provides a simple technical solution in patients requiring PFO closure when femoral venous access is not available. The classic jugular venous approach to PFO closure has always been considered difficult, but we describe the use of the EBU guiding catheter and a 180 cm 0.025" stainless steel guidewire with coiled floppy tip, which enables relatively simple passage across the PFO and provides sufficient support. This technique can be performed safely under the guidance of X-ray and TTE and provides an additional option for patients who require percutaneous PFO closure without femoral or hepatic venous access.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author/s.

AUTHOR CONTRIBUTIONS

All authors contributed to this patient care, diagnosis and treatment, to the writing of this article, and approved the submitted version.

REFERENCES

1. Sader MA, De Moor M, Pomerantsev E, Palacios IF. Percutaneous transcatheter patent foramen ovale closure using the right internal jugular venous approach. *Catheter Cardiovasc Interv.* (2003) 60:536–9. doi: 10.1002/ccd.10702
2. Hascoet S, Fraisse A, Elbaz M. Successful percutaneous transcatheter patent foramen ovale closure through the right internal jugular vein using a steerable catheter. *Catheter Cardiovasc Interv.* (2013) 82:E598–602. doi: 10.1002/ccd.24868
3. Fanari Z, Hammami S, Hopkins JT. Successful percutaneous transcatheter patent foramen ovale closure through the right internal jugular vein using stiff Amplatzer catheter with a reshaped tip. *Del Med J.* (2016) 88:238–41.
4. Mas JL, Derumeaux G, Guillon B, Massardier E, Hosseini H, Mechtouff L, et al. Patent foramen ovale closure or anticoagulation vs. antiplatelets after stroke. *N Engl J Med*. (2017) 377:1011–21. doi: 10.1056/NEJMoa1705915
5. Søndergaard L, Kasner SE, Rhodes JF, Andersen G, Iversen HK, Nielsen-Kudsk JE, et al. Patent foramen ovale clocsure or antiplatelet therapy for cryptogenic stroke. *N Engl J Med*. (2017) 377:1033–42. doi: 10.1056/NEJMoa1707404
6. Saver JL, Carroll JD, Thaler DE, Smalling RW, MacDonald LA, Marks DS, et al. Long-term outcomes of patent foramen ovale closure or medical therapy after stroke. *N Engl J Med*. (2017) 377:1022–32. doi: 10.1056/NEJMoa1610057
7. Lee PH, Song JK, Kim JS, Heo R, Lee S, Kim DH, et al. Cryptogenic stroke and high-risk patent foramen ovale: the DEFENSE-PFO trial. *J Am Coll Cardiol.* (2018) 71:2335–42. doi: 10.1016/j.jacc.2018.02.046
8. Alkhouli M, Campsey DM, Higgins L, Badhwar V, Diab A, Sengupta PP. Transcatheter closure of a sinus venosus atrial septal defect via transhepatic access. *JACC Cardiovasc Interv.* (2018) 11:e113–5. doi: 10.1016/j.jcin.2018.02.021
9. Hussain J, Strumpf R, Ghandforoush A, Jamal A, Diethrich E. Transhepatic approach to closure of patent foramen ovale: report of 2 cases in adults. *Tex Heart Inst J.* (2010) 37:553–6.
10. Jolly N, Dhar G, Amin Z. Transhepatic closure of patent foramen ovale. *Catheter Cardiovasc Interv.* (2010) 75:56–9. doi: 10.1002/ccd.22193
11. Ohta M, Hashizume M, Kawanaka H, Akazawa K, Ueno K, Tomikawa M, et al. Complications of percutaneous transhepatic catheterization of the portal venous system in patients with portal hypertension. *J Gastroenterol Hepatol.* (1996) 11:630–4. doi: 10.1111/j.1440-1746.1996.tb00305.x

12. Qintar M, Villablanca P, Lee J, Wang DD, Frisoli T, O'Neill B, et al. Patent foramen ovale closure with vena cava thrombus: you need an arm and a neck! *Clin Case Rep*. (2021) 9:e03884. doi: 10.1002/ccr3.3884

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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CASE REPORT
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Case Report: Challenging Treatment of an AorticParavalvular Leak: How We Avoided Interference With Mechanical Valve Function?

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Onorato EM, Vercellino M, Costante A and Bartorelli AL (2022) Case Report: Challenging Treatment of an AorticParavalvular Leak: How We Avoided Interference With Mechanical Valve Function? Front. Cardiovasc. Med. 9:839159. doi: 10.3389/fcvm.2022.839159

Background: Aortic paravalvular leak (APVL) after surgical valve replacement (AVR) is an ominous complication with a high risk of morbidity and mortality. Approximately 1–5% of PVLs can lead to serious clinical consequences, including congestive heart failure and/or hemolytic anemia.

Case Summary: A 69-year-old man with multiple comorbidities underwent surgical replacement of the aortic valve with a mechanical tilting disc prosthetic valve (Medtronic Starlight 27 mm). Several years later, recurrent episodes of congestive heart failure and hemolytic anemia developed due to a large crescent-shaped aortic PVL located at non coronary cusp (NCC) 9–12 o'clock, with moderate-to-severe regurgitation. The patient was deemed at prohibitive surgical risk due to significant multiple comorbidities and a transcatheter PVL closure (TPVLc) was planned. The huge PVL was partially closed by a first specifically designed paravalvular leak device (PLD). The procedure was complicated by transient interference of the second PLD with mechanical prosthetic valve function. This issue has however been solved with correct manipulation, orientation and downsizing of the second device implanted. At 3-month and 13-month follow-up, the patient showed a relevant clinical improvement and good quality of life. 2D TTE color Doppler confirmed the stable position of the two PLDs with trace residual leak.

Discussion: Surgical redo has been considered the treatment of choice for symptomatic patients with PVLs. Notwithstanding, TPVLc is a less invasive alternative, particularly in patients at high surgical risk in whom early diagnosis and prompt interventional treatment are crucial for improving expectancy and quality of life. Dedicated devices, appropriate procedural techniques, and the close interaction between imaging modalities, allowed to deal successfully with a challenging case of severe symptomatic aortic PVL.

Keywords: paravalvular leak, aortic valve, transcatheter closure, hemolytic anemia, heart failure

INTRODUCTION

Paravalvular leaks occur in patients who have undergone surgical valve replacement, with an incidence of 2–10% in the aortic position. Although most defects are small and asymptomatic, clinical manifestation of heart failure and hemolysis can be demonstrated in 1%-3% of patients, which leads to worst outcome and justify an interventional approach (1).

For patients with symptomatic PVL, surgical repair is the traditional treatment approach. Surgical results have shown to significantly improve patient outcomes at 5 and 10 years compared to medical treatment alone (2), despite the significant mortality associated with re-operation (3).

TPVLc is a less invasive treatment option, particularly in patients with prohibitive risk for redo surgery in whom it may represent a first-line treatment.

The safety and feasibility of TPVLc has been confirmed in several registries and a meta-analysis (4, 5). Overall technical success rate was 89.7%, being comparable between patients treated for mitral or aortic valve PVLs (92.6 vs. 83.3%, respectively) (6).

Nevertheless, catheter-based interventions are associated with higher rates of residual leaks in part due to complex anatomies, size and number of defects and to off-label use of devices designed for other applications.

CASE PRESENTATION

A 69-year-old male patient was admitted for recurrent episodes of congestive heart failure and hemolytic anemia. He has a past medical history of rheumatoid arthritis, Sjogren syndrome, moderate kidney disease (stage 3), chronic obstructive pulmonary disease, fronto-parietal left ischemic stroke with right side hemiparesis and rheumatic heart disease status following aortic valve replacement in 2005 with a mechanical tilting disc aortic prosthetic valve (Medtronic Starlight 27 mm). In the recent years, recurrent hospital admissions for heart failure and symptomatic hemolytic anemia requiring multiple red blood cells transfusions concomitantly occurred. The patient was in New York Heart Association (NYHA) class III under optimal medical therapy (OMT) (furosemide 50 mg/day, spironolactone 25 mg/day, bisoprolol 2.5 mg/day, ACE inhibitors 20 mg/day, aspirin 100 mg/day, statin 20 mg/day). Frequent episodes of dyspnea at rest with orthopnea (NYHA class IV) occurred during hemolysis, documented by low haptoglobin level and lactate dehydrogenase >500 UI/L. After red blood cell transfusions, the clinical and hemodynamic conditions improved significantly each time. Electrocardiogram showed biatrial enlargement, left bundle branch block and frequent ventricular ectopic beats. During each episode of congestive heart failure, chest X-ray revealed signs of increased pulmonary flow, dilatation of left-sided chambers as well as enlargement of pulmonary artery and its branches. Baseline

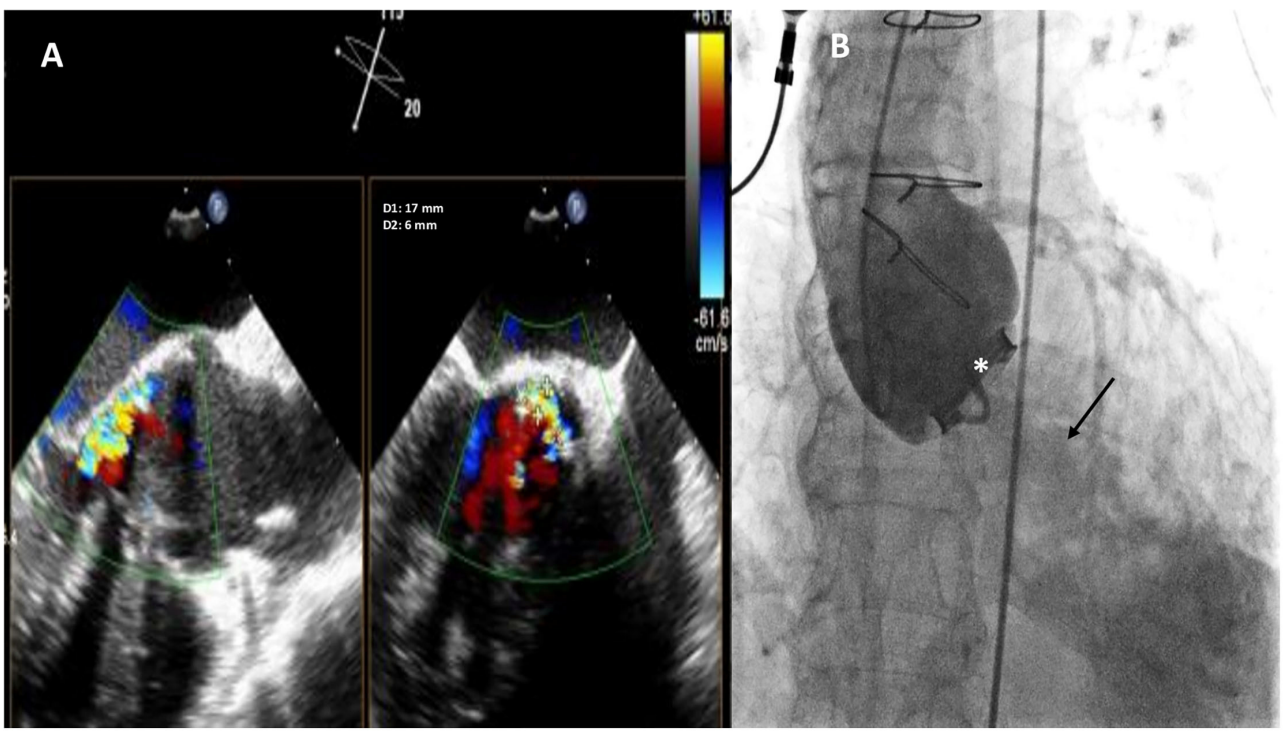

FIGURE 1 | Baseline two-dimensional (2D) X-plane imaging Transoeophageal Echocardiogram (TEE) color Doppler **(A)** and ascending aorta angiography **(B)** showing the presence of a huge crescent-shaped 17 × 6 mm aortic PVL located at non coronary cusp (NCC) 9-12 o'clock, with moderate-to-severe regurgitation. Fluoro-angiographic image confirming the stable position of the mechanical tilting disc aortic prosthetic valve (white asterisk) with moderate regurgitation (black arrow).

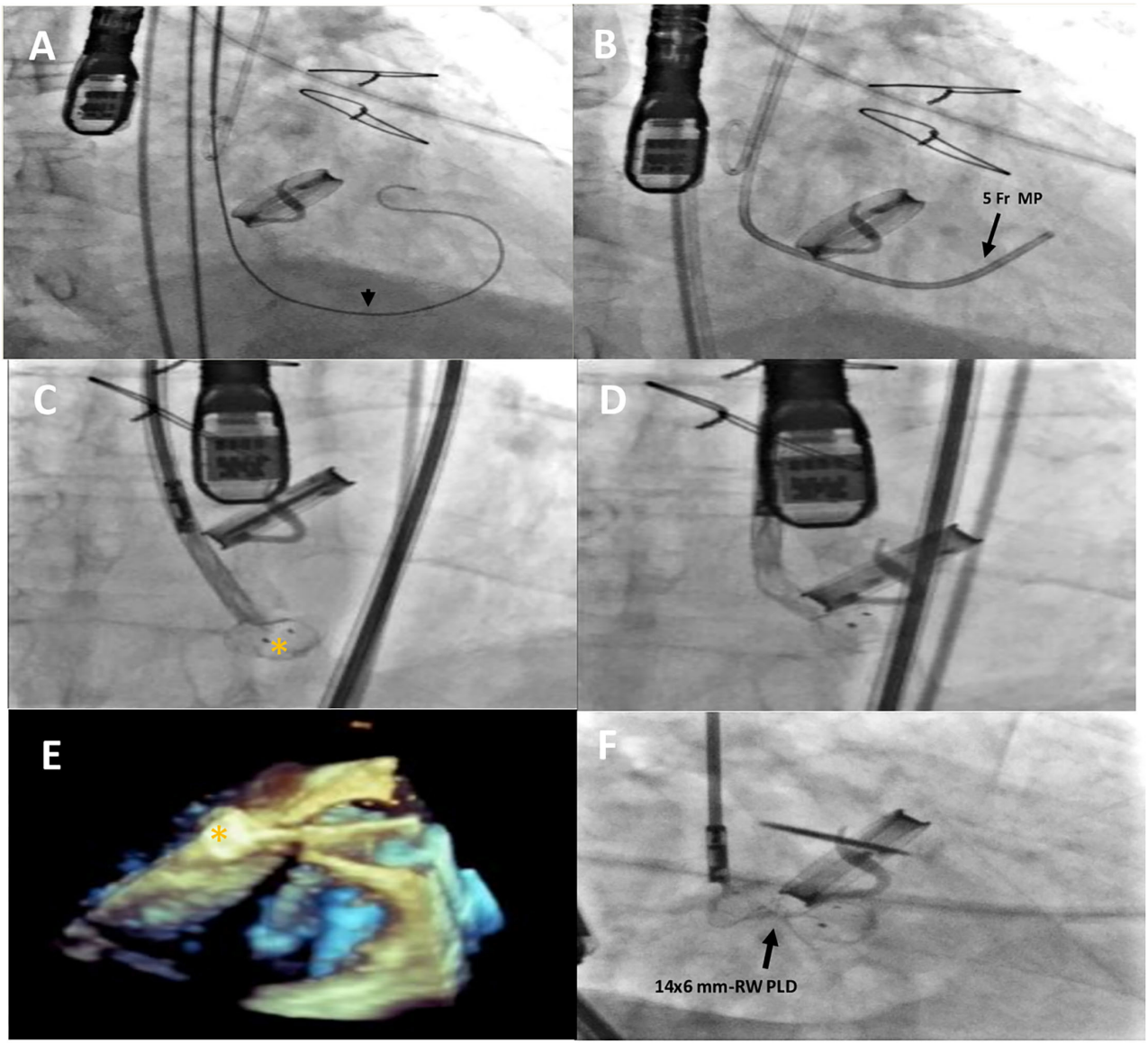

FIGURE 2 | Intraprocedural fluoro-angiographic and real-time 3D TEE procedural steps showing the guide wire **(A)** (black arrowhead) across the leakage with the distal soft tip in the left ventricle; the 5- Fr multipurpose catheter in the LV **(B)**; the distal disc device opening (orange asterisk) **(C–E)** and the 14 × 6 mm rectangular waist PLD correctly positioned and still anchored to the delivery cable **(F)**. MP, multipurpose catheter; PLD, Occlutech Parvalvular Leak Device; RW, rectangular waist.

two-dimensional (2D) Transthoracic/Transesophageal color Doppler Echocardiography (TTE/TEE) showed severe left ventricle (LV) dilatation (LV end-diastolic volume of 216 ml) with ejection fraction of 50% and confirmed the presence of a large 17 × 6 mm crescent-shaped aortic PVL located at non coronary cusp (NCC) 9–12 o'clock, with grade 4–5 aortic PVL regurgitation (PHT< 200 msec; vena contracta width = 10 mm) (7). Coronary angiography excluded coronary artery disease.

The risk stratification according to Society of Thoracic Surgeons (STS) score demonstrated an increased risk of long-term mortality (7%). According to 2014 AHA/ACC guidelines, TPVLc was a reasonable therapeutic option (class IIa, Level of Evidence B) in centers with expertise in symptomatic patients at high risk for surgical reintervention or with contraindication to reoperation (8). After heart team discussion, the decision to proceed with TPVLc was confirmed. Written informed consent, after explanation, was obtained from the patient. The procedure was performed under TEE and fluoroscopic guidance using general anesthesia. Prevention of contrast induced nephropathy was accomplished with normal saline 0.9% infusion (40 ml/h 12 h before and after procedure). Both left and right femoral arteries were cannulated. 4–5 degree paravalvular aortic regurgitation was demonstrated by 2D TEE color Doppler and by ascending

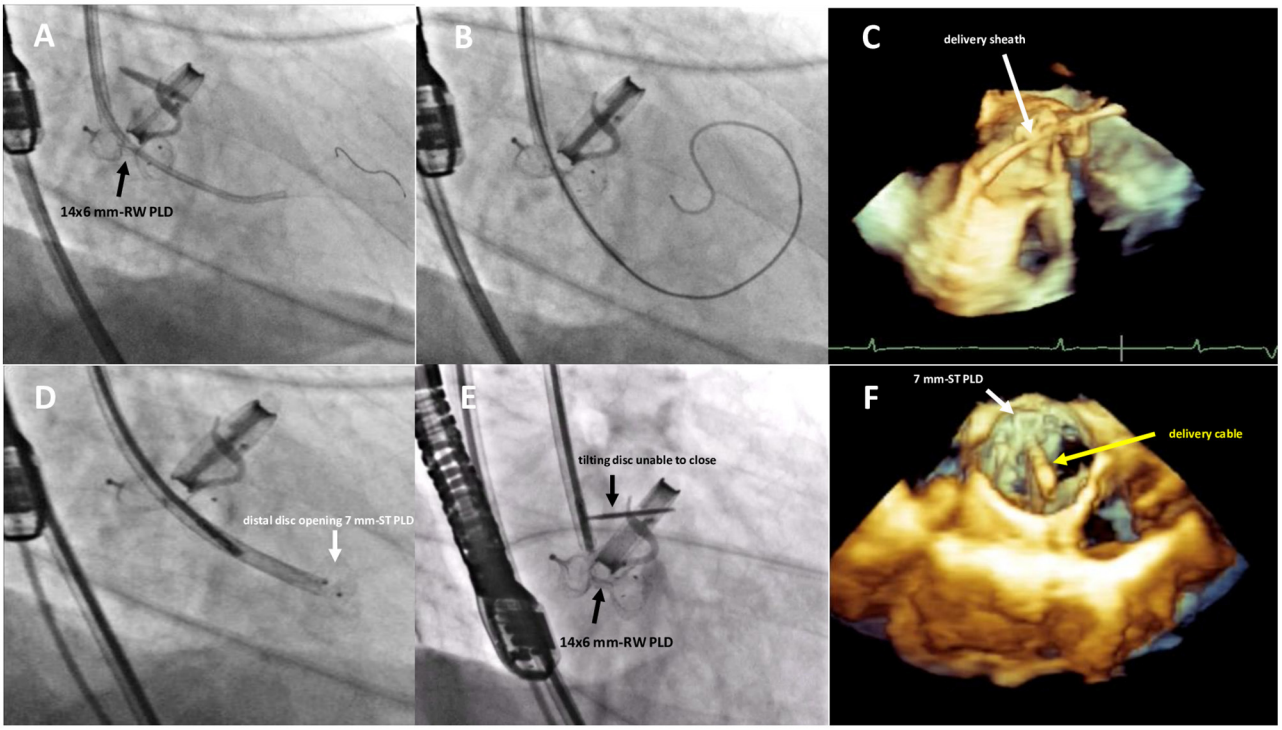

FIGURE 3 | Fluoro-angiographic and real-time 3D TEE procedural steps showing the crossing of the residual leakage in close proximity of the already implanted 14 × 6 mm rectangular waist PLD **(A–C)**, the distal disc opening of the 7 mm square twist PLD **(D)** still anchored to the delivery cable and the inference with the tilting disc of the mechanical aortic valve **(E,F)**. RW, rectangular waist; ST, square twist.

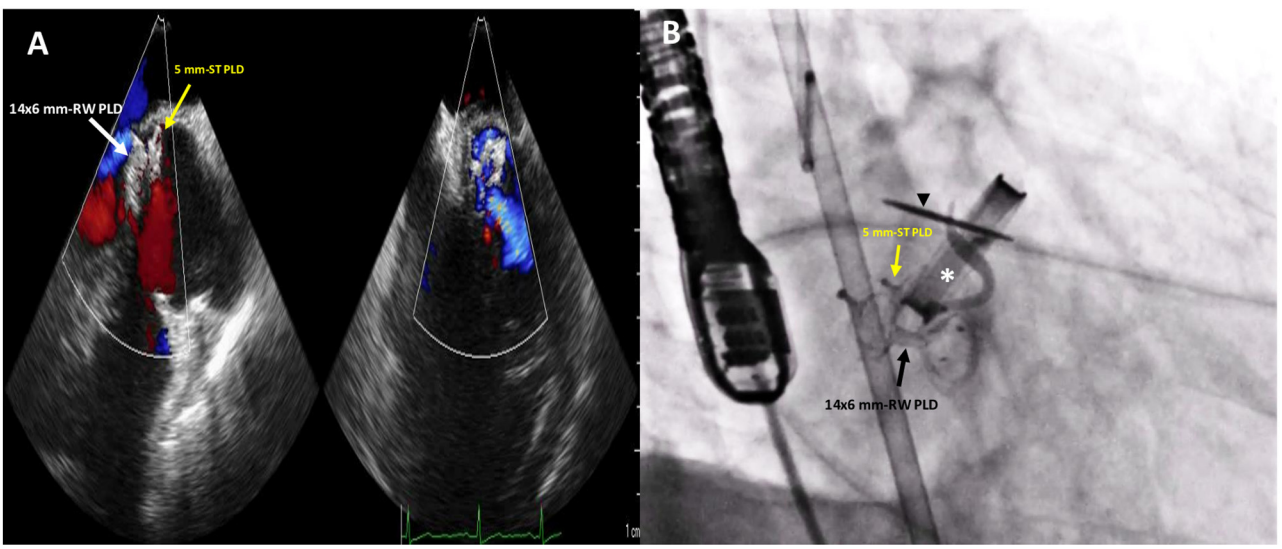

FIGURE 4 | Post-procedure 2D TTE color Doppler **(A)** and fluoro-angiographic **(B)** images showing the correct and stable position of the two PLDs not impinging on the tilting disc (black arrowhead) of the mechanical aortic valve (white asterisk) with only trivial residual regurgitant jet.

aortography with a 6-Fr pigtail angiographic catheter introduced from the left femoral artery (**Figure 1**). With the support of a 5-Fr multipurpose catheter (MP), introduced retrogradely from the right femoral artery, we easily advanced and passed through the huge NCC leakage a hydrophilic Terumo 0.035 inch guide wire, then replaced by a super stiff guidewire 0.035-inch

260 cm in lenght (Boston Scientific) that was positioned in the distal LV. The stronger support allowed progression of the 10-Fr delivery sheath into the LV. To accomplish the semilunar PVL dimensions (17 × 6 mm) with a height of 7 mm, a specifically designed 14 × 6 mm rectangular waist Paravalvular Leak Device (PLD, Occlutech, Helsingborg, Sweden) with a 24 mm distal disc, a 22 mm proximal disc and a 3 mm-height waist type of connection between the two disc was chosen and deployed (**Figure 2**). Intraprocedural 2D TEE color Doppler showed a persisting regurgitant jet via a 10 × 4 mm residual peri-device leakage probably due to the distorted "mushroom" spherical configuration of the two device disc and to elongated (up to 7 mm) shape of the connecting waist to fit the height of the original PVL (**Supplementary Figure 1**), hindering the apposition of the fabric within the disc. At this point, with the first PLD still anchored, the residual peri-device leak was crossed again with a hydrophilic guide wire, allowing the progression of a 5-Fr MP. A 0.035-inch wire 260 cm super stiff guidewire was advanced distally via the MP into the LV. The same 10-Fr delivery sheath was advanced over this exchange stiff wire allowing the progression of a second device, a 7 mm square twist PLD. After its opening, a massive intra-prosthetic aortic regurgitation was observed by the TEE monitoring. The angiogram revealed an acute interference between the 7 mm square twist PLD and the prosthetic valve, showing the tilting disc unable to close completely. Therefore, the 7 mm square twist PLD was then retrieved in the sheath and replaced by an undersized 5 mm square twist PLD that was implanted effectively without interfering anymore with the tilting disc of the mechanical aortic valve (**Figure 3**). Post-procedure echocardiographic and angiographic controls confirmed the effective PVL closure with only trivial residual regurgitant jet (**Figure 4**). The total volume of contrast medium was 80 cc. Peak creatinine level was 2.12 mg/dl 48 h after the procedure and 1.6 mg/dl at dismission, leading to the diagnosis of stage 1 acute kidney injury. The postoperative course was uneventful, and the patient was discharged on the eighth post-procedure day. At three-month and thirteen-month follow-up, he showed a relevant clinical improvement with a stable NYHA II class and good quality of life. No further hospitalizations and blood transfusions were needed. 2D TTE color Doppler confirmed the stable position of the two PLDs with trace residual leak (**Supplementary Figure 2**).

DISCUSSION

Comparison between surgical and percutaneous treatment have shown that technical success was higher with surgery [96.7 vs. 72.1%, odds ratio (OR) 9.7, $p < 0.001$] but at the cost of higher 30-day mortality (8.6 vs 6.8%, OR 1.90, $p < 0.001$), a trend toward higher stroke (3.3 vs. 1.4%, OR 1.94, $p = 0.069$), and longer hospitalizations. However, surgery was associated with similar 1-year mortality (17.3 vs. 17.2%, OR 1.07, $p = 0.67$), reoperation (9.1 vs. 9.9%, OR 0.72, $p = 0.1$), readmission for heart failure (13.3 vs. 26.4%, OR 0.51, $p = 0.29$), and improvement in New York Heart Association classification (67.4 vs. 56%, OR 1.37, $p = 0.74$), compared with percutaneous closure (9). Furthermore, PVL recurrence after first redo surgery has been reported to be 13% and increases further to 35% after second redo surgery (10). Reintervention rates were similar (11.3 vs. 17.2% in the percutaneous and surgical groups, respectively; $p = 0.10$), with the majority of reinterventions in the percutaneous group occurring early because of residual leak or persistent hemolysis. After risk adjustment, there was no significant difference in long-term survival between patients who underwent surgical vs. transcatheter treatment of PVLs (10).

TPVLc is a reasonable alternative to surgical intervention but it might be considered in patients with severe prosthetic paravalvular regurgitation with intractable hemolysis or NYHA class III or IV symptoms, who are at high or prohibitive surgical risk and have anatomical features suitable for percutaneous repair (11).

The efficacy of TPVL closure in reducing heart failure symptoms and long-term mortality has been demonstrated in multiple studies (12). Conversely, an incomplete TPVL closure could represent a very important limitation of this procedure, causing a worsening of hemolysis and NYHA class.

Undoubtedly, TPVL closure remains a complex, technically demanding and time-consuming procedure that requires not only careful patient selection and preprocedural multiple imaging modalities in order to visualize the complex 3D relationships of intracardiac structures. Additionally, it requires an active and strong collaboration of a skilled interventional team (interventional and imaging cardiologists, cardiac computed tomography radiologists and cardiac surgeons when needed).

Efforts are needed to develop new specifically designed devices to further improve clinical outcomes of this particular subset of high-risk patients with multiple comorbidities (13).

Even using a specific designed device, potential complications such as device malpositioning and interference between PLD and mechanical prosthetic valve may occur as demonstrated by our case. However, awareness of the pros and cons of each device, correct handling integrated with the echocardiographic imaging and good understanding of some procedural tips and tricks, could improve significantly the procedural success and clinical outcomes.

CONCLUSION

Our case demonstrates that even though TPVLc with a specifically designed device is a viable therapeutic alternative to surgical repair with high technical success rate, a close collaboration of a skilled interventional team is key for success, avoiding potential procedural complications such as interference with mechanical valve function and incomplete leakage closure that may increase NYHA class, the magnitude of hemolysis and the need for hemolysis-related blood transfusions.

AUTHOR'S NOTE

This paper was the original work of the authors who have all seen and approved of the paper and authorship. The article has not been published elsewhere and is not under consideration in any other journals.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/**Supplementary Material**, further inquiries can be directed to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Centro Cardiologico Monzino, IRCC, Milan, Italy. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

EO: conceptualization, writing—original draft preparation, visualization, and validation. MV and EO: methodology and data curation. AC: software. AB and EO: formal analysis, writing—review and editing, supervision, and investigation. All authors have read and agreed to the published version of the manuscript.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fcvm.2022.839159/full#supplementary-material

Supplementary Figure 1 | Post-procedure 2D X-plane **(A)** and short axis view **(B)** TEE color Doppler showing a regurgitant jet through a 10 × 4 mm residual peri-device leakage. RW, rectangular waist; PLD, Occlutech Parvalvular Leak Device.

Supplementary Figure 2 | Three-month follow-up 2D TTE **(A)** color Doppler **(B)** confirmed the stable position of the two PLDs with trace residual leak. PLD, Occlutech Paravalvular Leak Device.

REFERENCES

1. Kliger C, Eiros R, Isasti G, Einhorn B, Jelnin V, Cohen H, et al. Review of surgical prosthetic paravalvular leaks: diagnosis and catheter-based closure. *Eur Heart J.* (2013) 34:638–48. doi: 10.1093/eurheartj/ehs347
2. Genoni M, Franzen D, Vogt P, Seifert B, Jenni R, Kunzli A, et al. Paravalvular leakage after mitral valve replacement: improved long-term survival with aggressive surgery? *Eur J Cardiothorac Surg.* (2000) 17:14–9. doi: 10.1016/S1010-7940(99)00358-9
3. Emery RW, Krogh CC, McAdams S, Emery AM, Holter AR. Long-term follow up of patients undergoing reoperative surgery with aortic or mitral valve replacement using a St Jude medical prosthesis. *J Heart Valve Dis* (2010)19:473–84.
4. Cruz-Gonzales I, Rama-Merchan JC, Calvert P, Rodriguez-Collado J, Barreiro-Perez M, Martin-Moreiras J. et al. Percutaneous Closure of Paravalvular Leaks: A Systematic Review. *J Interven Cardiol.* (2016) 29:382–92. doi: 10.1111/joic.12295
5. Millán X, Skaf S, Joseph L, Ruiz C, García E, Smolka G, et al. Transcatheter reduction of paravalvular leaks: a systematic review and meta-analysis. *Can J Cardiol.* (2015) 31:260–9. doi: 10.1016/j.cjca.2014.12.012
6. Kalogeras K, Ntalekou K, Aggeli K, Moldovan C, Katsianos E, Kalantzis C, et al. Transcatheter closure of paravalvular leak: multicenter experience and follow-up. *Hellenic J Cardiol.* (2021) 62:416–22. doi: 10.1016/j.hjc.2021.02.002
7. Ruiz CE, Hahn RT, Berrebi A, Borer JS, Cutlip DE, Fontana G, et al. Clinical trial principles and endpoint definitions for paravalvular leaks in surgical prosthesis: an expert statement. *J Am Coll Cardiol.* (2017) 69:2067–87. doi: 10.1016/j.jacc.2017.02.038
8. Nishimura RA, Otto CM, Bonow RO, Carabello BA, Erwin 3rd JP, Guyton RA, et al. 2014 AHA/ACC guideline for the management of patients with valvular heart disease: Executive summary: a report of the American College of Cardiology/American Heart Association Task Force on Practice Guidelines. *J Am Coll Cardiol.* (2014) 63:2438–88. doi: 10.1161/CIR.0000000000000029
9. Busu T, Alqahtani F, Badhwar V, Cook CC, Rihal CS, Alkhouli M. Meta-analysis comparing transcatheter and surgical treatments of paravalvular leaks. *Am J Cardiol.* (2018) 122:302–9. doi: 10.1016/j.amjcard.2018.03.360
10. Akins CW, Bitondo JM, Hilgenberg AD, Vlahakes GJ, Madsen JC, MacGillivray TE. Early and late results of the surgical correction of cardiac prosthetic paravalvular leaks. *J Heart Valve Dis.* (2005) 14:792–9.
11. Otto CM, Nishimura RA, Bonow RO, et al. 2020 ACC/AHA guideline for the management of patients with valvular heart disease. *J Am Coll Cardiol.* (2020) 77:e25–e197. doi: 10.1016/j.jacc.2020.11.018
12. Calvert PA, Northridge DB, Malik IS, Shapiro L, Ludman P, Qureshi SA, et al. Percutaneous device closure of paravalvular leak: combined experience from the United Kingdom and Ireland. *Circulation.* (2016) 134:934–44. doi: 10.1161/CIR.0000000000000466
13. Loseph TA, Lane CE, Fender EA, Zack CJ, Rihal CS. Catheter-based closure of aortic and mitral paravalvular leaks: existing techniques and new frontiers. *Expert Rev Med Devices.* (2018) 15:653–63. doi: 10.1080/17434440.2018.1514257

Conflict of Interest: EO is a consultant for Occlutech, manufacturer of the device.

The remaining authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Case report: Transcatheter pulmonary valve-in-valve implantation in a deteriorated self-expandable valve caused by infective endocarditis

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Background: Infective endocarditis is a complication with high mortality in patients with congenital heart disease, particularly for those with bioprosthetic valve.

Case summary: We report a case of a 54-year-old female with a history of tetralogy of Fallot who had been surgically repaired using a transannular patch due to severe pulmonary insufficiency with right heart enlargement and presented with worsening dyspnea. She had received transcatheter pulmonary valve implantation (TPVI) 5 years ago. Unfortunately, bioprosthesis-associated infective endocarditis occurred due to dental caries. Given persistent antibiotic medication, she became clinically stable with prosthesis functional recovery. However, dysfunctional bioprosthesis was still detected 3 years later, which was successfully treated by valve-in-valve TPVI with the help of modified buddy wire technique. At a 12-month follow-up after valve-in-valve TPVI, she was completely recovered with improved symptoms of heart failure.

Conclusion: This is the first report of valve-in-valve TPVI of a self-expandable valve in a degenerated self-expandable valve. The case highlights increased surveillance for infective endocarditis of transcatheter pulmonary valve should be emphasized. Subsequent valve-in-valve TPVI is an effective treatment for valve failure in defined conditions improving the hemodynamics.

KEYWORDS

degenerated bioprosthesis, infective endocarditis, tetralogy of Fallot, transcatheter pulmonary valve implantation, valve-in-valve (VIV)

Introduction

Pulmonary regurgitation is a common late consequence of surgical repair of tetralogy of Fallot (TOF) (1, 2). European society of cardiology guidelines on the management of adult congenital heart disease recommend transcatheter pulmonary valve implantation (TPVI) in symptomatic patients affected by severe pulmonary regurgitation (3). Given the relatively low risk of procedural complications, TPVI may be an optimal option in selective cases. Although TPVI has been reported to be a feasible procedure

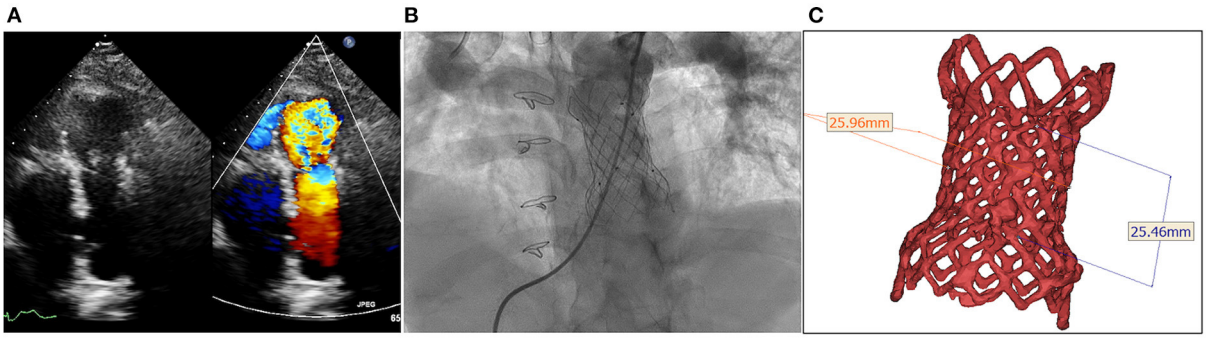

FIGURE 1
Degenerated bioprosthesis with complete frame. **(A)** Transthoracic echocardiography shows severe pulmonary regurgitation; **(B)** Angiography reveals severe pulmonary insufficiency; **(C)** The annular size calculated on computed tomography.

with successful implantation rate >95% (4, 5), it remains challenging and technically demanding in some special scenario.

In this report, we present a treatment modality for self-expandable valve failure caused by infective endocarditis (IE), using valve-in-valve TPVI with the assistance of modified buddy wire technique.

Case description

A 54-year-old female with a history of TOF was admitted to our hospital with complains of exertional dyspnea and chest distress. She underwent surgery for TOF with transannular patching of right ventricular outflow tract (RVOT) at the age of 18. Five years ago, she received TPVI, using a P26-25 mm (diameter-length) self-expandable Venus P-valve (Venus Medtech, Hangzhou, China) due to severe pulmonary regurgitation with right heart enlargement. Unfortunately, bioprosthesis-associated IE occurred due to dental caries 6 months after TPVI. Transthoracic echocardiography (TTE) showed vegetation attached on the prosthetic valve and blood culture indicated streptococcus viridians. At the end of 6 weeks of intravenous penicillin and gentamycin, her blood cultures sterilized, her vegetation size was reduced, and she achieved prosthesis functional recovery (6). Then, she received dental caries extraction under antibiotic prophylaxis with intravenous penicillin 80 million units/day for 3 days. During close follow-up, she had not got relapse of IE. However, dysfunctional bioprosthesis was still detected by TTE with thicken leaflet, severe pulmonary regurgitation 3 years later (Figure 1A; Supplementary Video 1), moderate to severe tricuspid insufficiency and right heart dilation. Considering the relatively low risk of procedural complications, a decision of valve-in-valve TPVI with a self-expandable valve was made.

The procedure was performed with fluoroscopic and TTE guidance under general anesthesia. Bilateral femoral veins were used, and right heart catheterization was first executed. Pulmonary artery pressure was 35/8 mmHg and right ventricle pressure was 50/8 mmHg. Severe pulmonary insufficiency appeared on angiography (Figure 1B; Supplementary Video 2). After pre-dilation with a 25 mm × 50 mm (diameter × length) balloon (Balt, Montmorency, France), systolic pressure gradient across the bioprosthesis decreased from 15 mmHg to 6 mmHg. Since the annular size of 25.9 mm calculated on computed tomography and three-dimension reconstruction (Figure 1C), a P28-25 mm (diameter-length) self-expandable Venus P-valve (Venus Medtech, Hangzhou, China) with 22-F delivery system was planned to be deployed. The process was beset with difficulties in advancing the delivery sheath. The buddy wire technique, which was employed with two Lunderquist wires, was attempted but the delivery system still failed to be advanced and positioned appropriately. Finally, the 14-F Cook sheath (Cook Medical, Bloomington, USA) was advanced over the second Lunderquist wire, which passed through the RVOT and placed in the distal of right pulmonary artery. The 14-F Cook sheath served as modified buddy wire providing extra support and straightening the vessel and the RVOT (Figure 2A). Maneuvering the 22-F Venus P delivery system alongside the 14-F Cook sheath aided in the advancement of the delivery system into the proximal end of left pulmonary. Prior to the deployment of the second pulmonary valve, the 14-F Cook sheath was pulled back into inferior vena cava. The new self-expanding valve was then successfully delivered (Figure 2B) and implanted. Post-dilation with a 25 mm × 50 mm (diameter × length) balloon (Balt, Montmorency, France) was performed to better shape for the stent strut and RVOT. Post-procedure angiography and TTE showed no pulmonary regurgitation

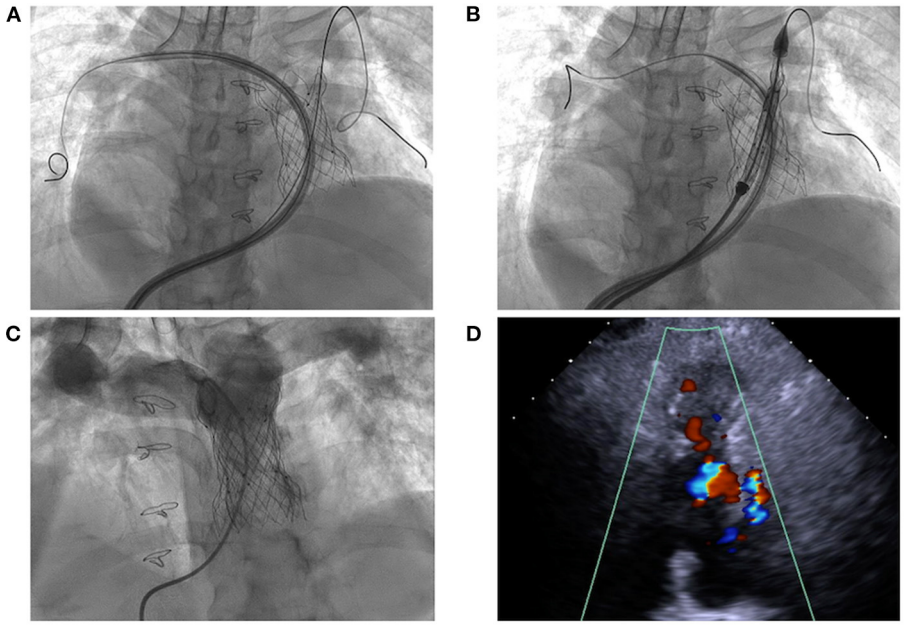

FIGURE 2
Transcatheter pulmonary valve-in-valve implantation for degenerated bioprosthesis. **(A,B)** The delivery system crossed the degenerated bioprostheses with the assistance of modified buddy wire; **(C,D)** The result of transcatheter valve-in-valve implantation with no pulmonary regurgitation.

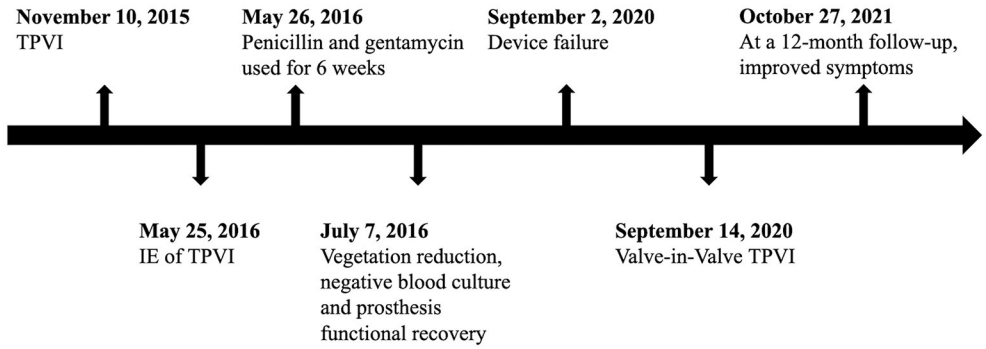

FIGURE 3
Timeline of events of the patient.

(Figures 2C,D, 3; Supplementary Videos 3, 4). Then pulmonary pressure was raised to 35/18 mmHg.

During hospitalization, the patient was given adequate and prolonged antibiotic prophylaxis (intravenous ceftriaxone 2 g/day and vancomycin 1 g/day for 1 week) to ensure no endocarditis relapse. She was recommended to take oral anticoagulant daily after discharge and reinforced follow-up was proposed to her as before. At 12-month follow-up, she was completely recovered from valve-in-valve TPVI with improved symptoms of heart failure (Figure 3). TTE was obtained and showed a persistently good valve function.

Discussion

TPVI is an effective treatment alternative to surgical pulmonary valve replacement for patients with right ventricular dysfunction after correction of TOF (7–10). IE of TPVI is a dreadful complication and associated with a relevant need of re-intervention (11). IE of Venus P valve with concomitant COVID-19 infection was also reported (12). In our case, the mechanism resulting in recurrence of pulmonary regurgitation after TPVI was IE, eventually leading to recurrence of pulmonary regurgitation and right

heart failure. Although transcatheter procedure could be the first reintervention, it was necessary to identify that IE was adequately treated, which was confirmed by no clinical sign and no vegetations on echocardiogram in this scenario.

This is the first report of valve-in-valve TPVI in a degenerated self-expandable pulmonary valve. Maneuvering the relatively rigid delivery system can be challenging in a dilated and tortuous right ventricle with severe tricuspid insufficiency, and it is difficult to advance the valve especially in a stented RVOT. Although several reported technical and therapeutic considerations remain to be resolved in difficult TPVI cases (13–15), the modified buddy wire technique including super stiff wire combined with long sheath to provide extra guide and support could be a bailout approach, which facilitates to maneuver the delivery system crossing the basal valve frame and deploy the new valve at accurate position. Albeit limited data were available regarding the hemodynamic efficacy and durability of such intervention, repeat TPVI was still an effective treatment for valve failure in defined conditions, improving by freedom from re-intervention, which further adds to the benefit of TPVI in the life-time management of RVOT dysfunction (16). During the procedure, the technique along with the wire and sheath can be used to place percutaneous valve in accurate position.

In conclusion, this case highlights reinforced surveillance for IE of transcatheter pulmonary valve should be emphasized. Subsequent valve-in-valve TPVR is an effective treatment for early bioprosthetic failure in defined conditions. Repeat TPVI is particularly challenging in case with self-expandable valve but is an effective treatment with the modified buddy wire technique to provide extra support for delivery system.

Data availability statement

The original contributions presented in the study are included in the article/Supplementary material, further inquiries can be directed to the corresponding author.

Ethics statement

The studies involving human participants were reviewed and approved by the Human Ethics Committee of Shanghai Chest Hospital, Shanghai Jiao Tong University. The patients/participants provided their written informed consent to participate in this study. Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

Author contributions

Y-JL, XP, CW, and BH contributed to the conception, design, and implement of the study. Y-JL and XP organized the data, performed the statistical analysis, and wrote the manuscript. All authors contributed to manuscript, read, and approved the submitted version for publication.

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Conflict of interest

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Publisher's note

Supplementary material

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SUPPLEMENTARY VIDEO S1
Thickened leaflet and severe pulmonary regurgitation on echocardiography.

SUPPLEMENTARY VIDEO S2
Severe pulmonary regurgitation on angiography.

SUPPLEMENTARY VIDEO S3
Post-procedure angiography showed no pulmonary regurgitation.

SUPPLEMENTARY VIDEO S4
No regurgitation after transcatheter valve-in-valve implantation on echocardiography.

References

1. Greutmann M, Ruperti J, Schwitz F, Haag N, Santos Lopes B, Meier L, et al. High variability of right ventricular volumes and function in adults with severe pulmonary regurgitation late after tetralogy of fallot repair. *Am J Cardiol.* (2022) 166:88–96. doi: 10.1016/j.amjcard.2021.11.022

2. Liu J, Jiang X, Peng B, Li S, Yan J, Wang Q, et al. Association of pulmonary valve morphology differences with outcomes in tetralogy of fallot repair with right ventricular outflow tract incision. *Front Cardiovasc Med.* (2021) 8:695876. doi: 10.3389/fcvm.2021.695876

3. Baumgartner H, De Backer J, Babu-Narayan SV, Budts W, Chessa M, Diller GP, et al. 2020 ESC Guidelines for the management of adult congenital heart disease. *Eur Heart J.* (2021) 42:563–645. doi: 10.15829/1560-4071-2021-4702

4. Sinha S, Aboulhosn J, Levi DS. Transcatheter pulmonary valve replacement in congenital heart disease. *Interv Cardiol Clin.* (2019) 8:59–71. doi: 10.1016/j.iccl.2018.08.006

5. Tannous P, Nugent A. Transcatheter pulmonary valve replacement in native and nonconduit right ventricle outflow tracts. *J Thorac Cardiovasc Surg.* (2021) 162:967–70. doi: 10.1016/j.jtcvs.2020.07.126

6. Wang C, Li YJ, Ma L, Pan X. Infective endocarditis in a patient with transcatheter pulmonary valve implantation. *Int Heart J.* (2019) 60:983–5. doi: 10.1536/ihj.18-497

7. Jones TK, McElhinney DB, Vincent JA, Hellenbrand WE, Cheatham JP, Berman DP, et al. Long-term outcomes after Melody transcatheter pulmonary valve replacement in the US investigational device exemption trial. *Circ Cardiovasc Interv.* (2022) 15:e010852. doi: 10.1161/CIRCINTERVENTIONS.121.010852

8. Lee SY, Kim GB, Kim SH, Jang SI, Choi JY, Kang IS, et al. Mid-term outcomes of the Pulsta transcatheter pulmonary valve for the native right ventricular outflow tract. *Catheter Cardiovasc Interv.* (2021) 98:E724–32. doi: 10.1002/ccd.29865

9. McElhinney DB, Zhang Y, Levi DS, Georgiev S, Biernacka EK, Goldstein BH, et al. Reintervention and survival after transcatheter pulmonary valve replacement. *J Am Coll Cardiol.* (2022) 79:18–32. doi: 10.1016/j.jacc.2021.10.031

10. Morgan G, Prachasilchai P, Promphan W, Rosenthal E, Sivakumar K, Kappanayil M, et al. Medium-term results of percutaneous pulmonary valve implantation using the Venus P-valve: international experience. *EuroIntervention.* (2019) 14:1363–70. doi: 10.4244/EIJ-D-18-00299

11. Bos D, De Wolf D, Cools B, Eyskens B, Hubrechts J, Boshoff D, et al. Infective endocarditis in patients after percutaneous pulmonary valve implantation with the stent-mounted bovine jugular vein valve: clinical experience and evaluation of the modified Duke criteria. *Int J Cardiol.* (2021) 323:40–6. doi: 10.1016/j.ijcard.2020.08.058

12. Chimoriya R, Awasthy N, Kumar G. COVID-19 infection with delayed presentation of infective endocarditis of the prosthetic pulmonary valve. *Cardiol Young.* (2021) 31:2045–7. doi: 10.1017/S1047951121002080

13. Baz Alonso JA, Fernandez GB, Barbeira SF, Romo AI, Zunzunegui JL, Jimenez-Diaz VA. Transcatheter pulmonary valve replacement with "Double-Barrel" stent-and-valve technique in a dilated right ventricular outflow tract. *JACC Cardiovasc Interv.* (2021) 14:e283–e4. doi: 10.1016/j.jcin.2021.08.015

14. Rudzinski PN, Kalinczuk L, Mintz GS, Demkow M. Large field-of-view intravascular ultrasound offering tomographic perspective online for accurate sizing during transcatheter pulmonary valve replacement. *Eur Heart J Cardiovasc Imaging.* (2022) jeac075. doi: 10.1093/ehjci/jeac075

15. Shah RR, Poommipanit P, Law MA, Amin Z. Anchor balloon, buddy wire, and wire and sheath techniques to deploy percutaneous pulmonary valves in tetralogy of fallot patients. *Catheter Cardiovasc Interv.* (2018) 92:915–20. doi: 10.1002/ccd.27022

16. Nordmeyer J, Coats L, Lurz P, Lee TY, Derrick G, Rees P, et al. Percutaneous pulmonary valve-in-valve implantation: a successful treatment concept for early device failure. *Eur Heart J.* (2008) 29:810–5. doi: 10.1093/eurheartj/ehn073

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Case report: Intrapericardial thrombus aspiration in early stage of pericardial thrombosis for cardiac tamponade complicating percutaneous left atrial appendage closure

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Introduction: Pericardial thrombosis that complicates pericardial bleeding is a life-threatening emergency in interventional cardiology, and surgery remains the only definitive treatment option. We report the first case of successful intrapericardial thrombus aspiration using a dedicated thrombus aspiration catheter in the early stage of pericardial thrombosis.

Case report: A 76-year-old woman with non-valve atrial fibrillation underwent percutaneous left atrial appendage (LAA) closure for secondary prevention of stroke. A 24-mm Watchman device was deployed under fluoroscopic guidance. Post-deployment angiography revealed LAA perforation, which led to the rapid onset of cardiac tamponade. Emergency pericardiocentesis was performed and the deep-seated device was redeployed at a more proximal position to seal the distal perforation. Pericardial bleeding was controlled after the drainage of 400 ml of blood. However, the patient re-developed cardiac tamponade following a period of stability. The patient was diagnosed with early-stage pericardial thrombosis based on echocardiographic findings of a hypoechoic effusion in the pericardial space. Thrombus aspiration using a pigtail catheter and long sheath failed; however, we performed successful intrapericardial thrombus aspiration using a dedicated thrombus aspiration catheter. We drained 120 ml of sludge-like blood, and the patient underwent successful conservative management without surgical intervention.

Conclusion: This case report highlights the potential usefulness of a percutaneous intrapericardial thrombus aspiration technique using a dedicated thrombus aspiration catheter in selected patients with early-stage pericardial thrombosis, as a less invasive alternative to cardiac surgery.

KEYWORDS

thrombus aspiration, pericardial thrombosis, cardiac tamponade, left atrial appendage closure, case report

Introduction

Cardiac tamponade is a potentially fatal complication in patients who undergo percutaneous cardiac interventions (1–3). Procedural-related cardiac or coronary perforations can cause rapid accumulation of blood in the pericardium and may precipitate a cardiac emergency (4). Emergency pericardiocentesis is a life-saving procedure in hemodynamically unstable patients with clinical evidence of cardiac tamponade (5).

Reportedly, the incidence of pericardial effusion that necessitates intervention was 4.8 and 1.9% in randomized left atrial appendage closure (LAAC) trials of PROTECT AF and PREVAIL, respectively (6, 7). More recent studies have reported a lower pericardial effusion rate of less than 1–2% (8, 9). Most intervention-induced perforations (approximately 88%) can be treated conservatively using pericardiocentesis, without surgical intervention (5). However, open surgical repair is necessary for patients with prolonged uncontrolled bleeding or pericardial thrombosis (10). The latter is a more fatal complication; pericardial thrombus significantly restricts the diastolic function of the heart (4) and cannot usually be aspirated using a pericardial drainage tube. We report a case of successful intrapericardial thrombus aspiration using a dedicated thrombus aspiration catheter during early-stage pericardial thrombosis, as a less invasive alternative to cardiac surgery.

Case

A 76-year-old woman with a history of non-valve atrial fibrillation (AF), hypertension, diabetes, prior ischemic stroke, a CHA_2DS_2-VASc score of 7, and a HAS-BLED score of 3 underwent percutaneous LAAC for secondary prevention of stroke.

The LAAC procedure was performed under local anesthesia, deep sedation, and fluoroscopic guidance. A decapolar catheter was inserted through the right femoral vein into the coronary sinus to guide transseptal puncture. Unfractionated heparin (100 U/kg) was administered immediately following transseptal puncture to achieve an activated clotting time of 330 s. The left atrial pressure was 33/15 mmHg. Left atrial appendage (LAA) angiography (**Figure 1A** and **Supplementary Video 1**) at the right anterior oblique 30° and caudal 20° view revealed ostial width of 19.5 mm and depth of 21.8 mm. A 24-mm Watchman device (Boston Scientific, MA, United States) was selected and then deployed under fluoroscopic guidance. Post-deployment angiography revealed brisk contrast extravasation into the pericardial space (**Figure 1B** and **Supplementary Video 2**). The patient rapidly developed cardiac tamponade, and blood pressure decreased from 131/72 to 78/35 mmHg.

We performed emergency pericardiocentesis via a subxiphoid approach under fluoroscopic guidance. A pigtail catheter was inserted into the pericardial cavity to drain blood, and the aspirated pericardial blood was immediately returned to the femoral vein via a sheath. Protamine (30 mg) was simultaneously administered to reverse heparin activity. The patient's systolic blood pressure returned to 95 mmHg after aspiration of 150 ml of blood. The device was retracted and redeployed at a more proximal position to effectively seal the LAA and distal perforation (**Figure 1C** and **Supplementary Video 3**). After confirmation of device stability and absence of residual peri-device leakage, the Watchman device was released (**Figure 1D** and **Supplementary Video 4**). Repeat aspiration was performed for an additional 15 min. The patient's vital signs stabilized, and blood pressure returned to 120/65 mmHg. The pericardial fluid was drained to dryness after aspiration of 400 ml of blood, and minimal pericardial fluid reaccumulation was observed.

Following 10 min observation, the patient's heart rate showed intermittent slowing with a decrease in blood pressure to 90/62 mmHg. A temporary pacing lead was placed into the right ventricular to maintain a ventricular rate of $>$ 60 beats per minute. Fluoroscopy revealed a near-normal-sized cardiac silhouette (**Figure 2A** and **Supplementary Video 5**), and minimal pericardial blood was drained after the insertion of a new pigtail catheter. The decrease in blood pressure was likely attributable to suspected pericardial thrombosis. Emergency echocardiography revealed a hypoechoic (rather than anechoic) effusion in the pericardial space (**Figure 3A**), suggestive of early pericardial thrombosis. The emergency surgical team was summoned to prepare for open chest surgery.

The patient's blood pressure was temporarily stable at approximately 90/60 mmHg; therefore, intrapericardial thrombus aspiration was attempted before surgery. However, thrombus aspiration failed using a pigtail catheter, which was replaced by an 8.5 F long sheath (SL1, Abbott, MN, United States). Unfortunately, thrombus aspiration through the sheath was also unsuccessful. Thereafter, we used a dedicated thrombus aspiration catheter. A 6F guiding catheter (Judkins R4.0, Medtronic, MN, United States) was inserted through the long sheath into the pericardial cavity via an angioplasty guidewire (BMW, 0.036 cm $\times$ 190 cm, Abbott, MN, United States), and a thrombus aspiration catheter (Thrombuster II, Kaneka Medical Products, Osaka, Japan) was advanced into the pericardial cavity via the guidewire. Following manipulation of the guiding catheter and guidewire, we could maneuver the thrombus aspiration catheter to successfully aspirate the thrombus from multiple sites across the pericardium (**Figures 2B,C** and **Supplementary Videos 6, 7**). Sludge-like blood (instead of a thrombus) was drained using the aspiration catheter (**Figure 2D**). We aspirated 120 ml of sludge-like blood after 10 min. The patient was hemodynamically stable, and blood pressure returned to

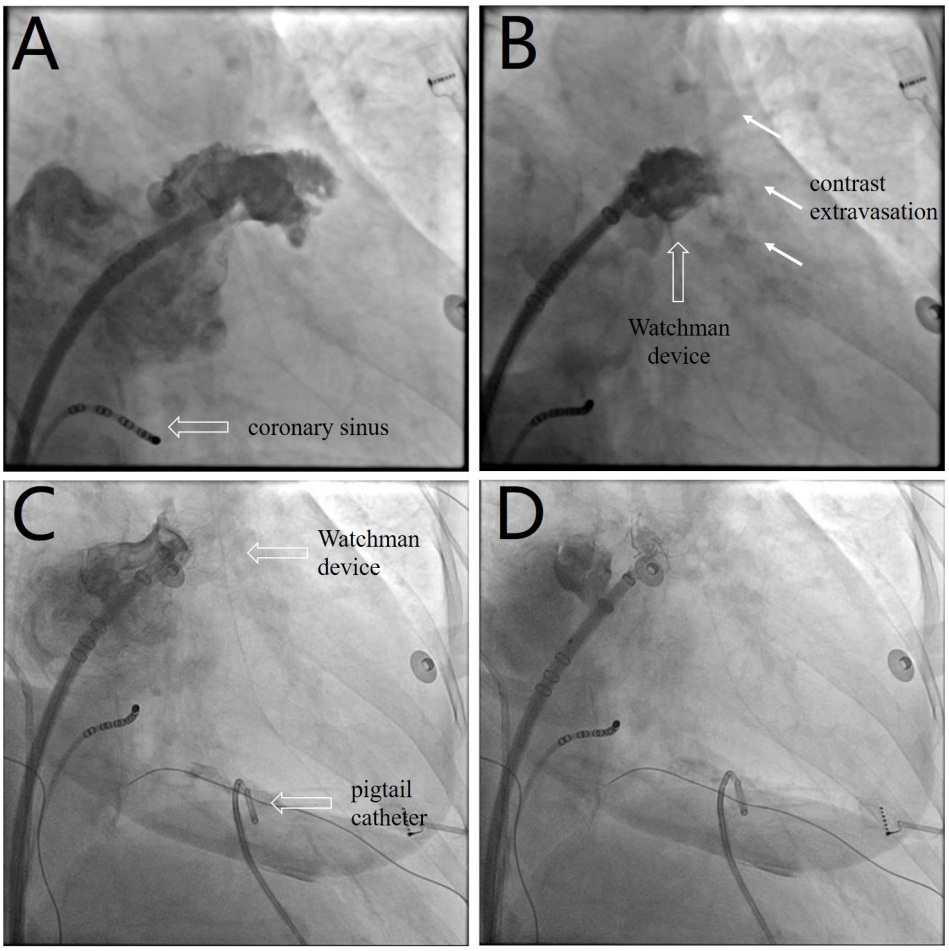

FIGURE 1
(A) Left atrial appendage angiography before device selection (**Supplementary Video 1**). **(B)** Post-deployment angiography shows brisk contrast extravasation (white arrow) into the pericardial space (**Supplementary Video 2**). **(C)** The device is redeployed at a more proximal position to seal the appendage and distal perforation (**Supplementary Video 3**). **(D)** The device is released after confirmation of device stability and the absence of residual peri-device leakage (**Supplementary Video 4**).

123/62 mmHg. Echocardiography revealed mild effusion and a round hyperechoic thrombus (2.5 cm × 1.7 cm) in the vicinity of the right ventricular apex (**Figure 3B**). A pigtail catheter was placed to monitor the pericardium, and the patient was transferred back to the ward.

The pigtail catheter was removed on the second postoperative day after echocardiography confirmed the absence of pericardial fluid reaccumulation. We observed shrinkage of the round thrombus, which appeared as a strip that measured 1.8 cm × 0.7 cm in size near the right ventricular apex (**Figure 3C** and **Supplementary Video 8**). Anticoagulation was re-initiated on the third postoperative day, and the patient was discharged on the fifth postoperative day. Echocardiography performed 2 weeks after discharge revealed no thrombus or pericardial effusion (**Figure 3D** and **Supplementary Video 9**), and the patient had no thromboembolic event or pericardial effusion during 1-year follow-up.

Discussion

Pericardial thrombosis is a life-threatening emergency, and therapeutic pericardiocentesis is challenging. We report the first case of successful intrapericardial thrombus aspiration using a dedicated thrombus aspiration catheter in the early stage of pericardiac thrombosis complicating LAAC, providing a percutaneous treatment of early pericardial thrombosis.

Percutaneous LAAC serves as an alternative to oral anticoagulation for stroke prevention in patients with non-valve AF by mechanical occlusion of the LAA. The LAA is a thin-walled structure (11); therefore, it is highly vulnerable to tears or perforation following manipulation by a closure device or catheter, which may lead to pericardial bleeding and potential cardiac tamponade. The rate of pericardial tamponade that complicates LAAC has decreased owing to the implementation of standardized operative procedures and greater accumulation

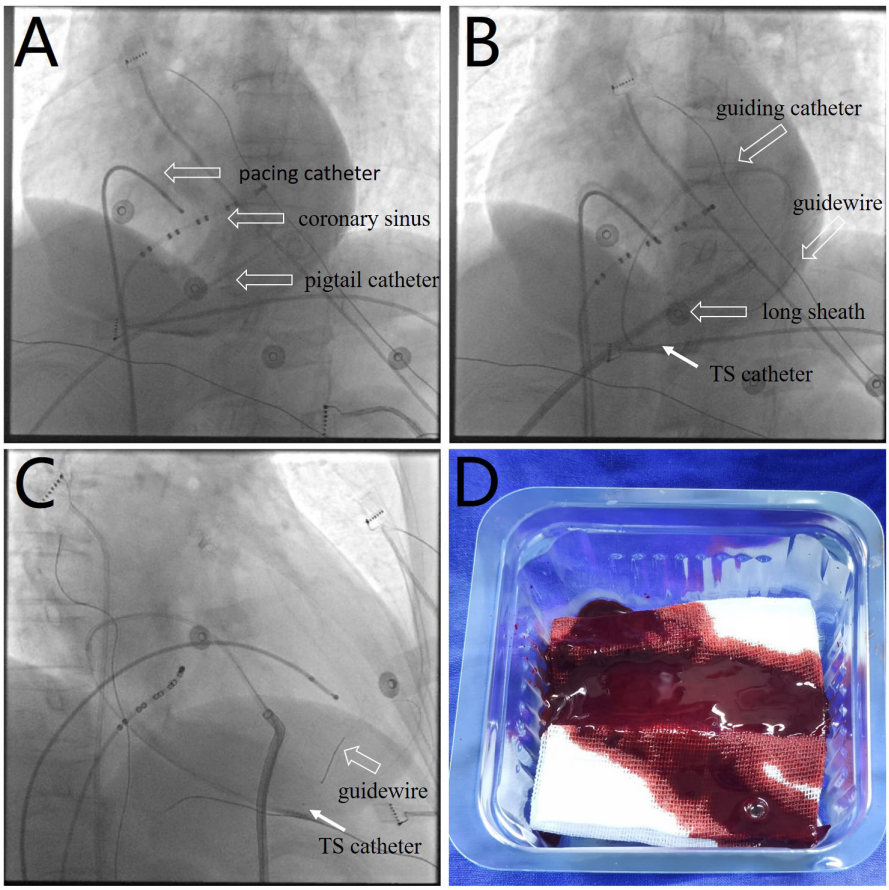

FIGURE 2
(A) Fluoroscopic image showing a near-normal sized cardiac silhouette (**Supplementary Video 5**), although the patient's blood pressure decreased to 90/62 mmHg. **(B,C)** Images showing the use of a thrombus aspiration catheter (white arrows) for thrombus aspiration from multiple pericardial sites with cautious manipulation of the guiding catheter and guidewire (**Supplementary Videos 6, 7**). **(D)** Image showing a sludge-like appearance of blood (as opposed to a formed thrombus) drained by the aspiration catheter. TS, thrombus aspiration.

of surgical experience (12); however, pericardial tamponade remains a serious complication of percutaneous LAAC.

Emergency pericardiocentesis is performed to relieve cardiac tamponade in patients with hemodynamic instability. Pericardiocentesis is an effective conservative treatment approach in most patients with pericardial tamponade that complicates LAAC (5). In our case, the closure device was deployed too deep and perforated the LAA, which precipitated pericardial tamponade. Emergency pericardiocentesis was performed to relieve cardiac tamponade. The deep deployed device was redeployed at a more proximal position to seal the distal perforation, and heparin was reversed. These measures effectively controlled pericardial bleeding.

However, the patient returned with cardiac tamponade again after a period of stability and was diagnosed with pericardial thrombosis, following echocardiographic evaluation. Surgical treatment is the only definitive therapeutic option available for pericardial thrombosis (13) and to date, no study has reported successful intrapericardial thrombus aspiration in such cases. Temporary stabilization of the patient's blood pressure facilitated intrapericardial thrombus aspiration under backup surgery. After the failure of the pigtail catheter and long sheath aspiration, intrapericardial thrombus aspiration was successful using the thrombus aspiration catheter (Thrombuster II), which is attributable to the following advantages offered by this novel approach. The Thrombuster II catheter with a large circular lumen and superior aspiration ability is dedicated for thrombus aspiration. Echocardiography revealed hypoechoic and not anechoic or hyperechoic effusion, which suggested early-stage pericardial thrombosis; incomplete thrombus formation enabled aspiration and the sludge-like quality of drained blood confirmed this finding.

Intra-procedural surveillance of transesophageal echocardiography (TEE) was not performed, which serves as a significant limitation of this study. TEE enables timely detection of pericardial effusion and thrombosis, and can outline the process from pericardial effusion to thrombus formation. Our case is quite unique because pericardial

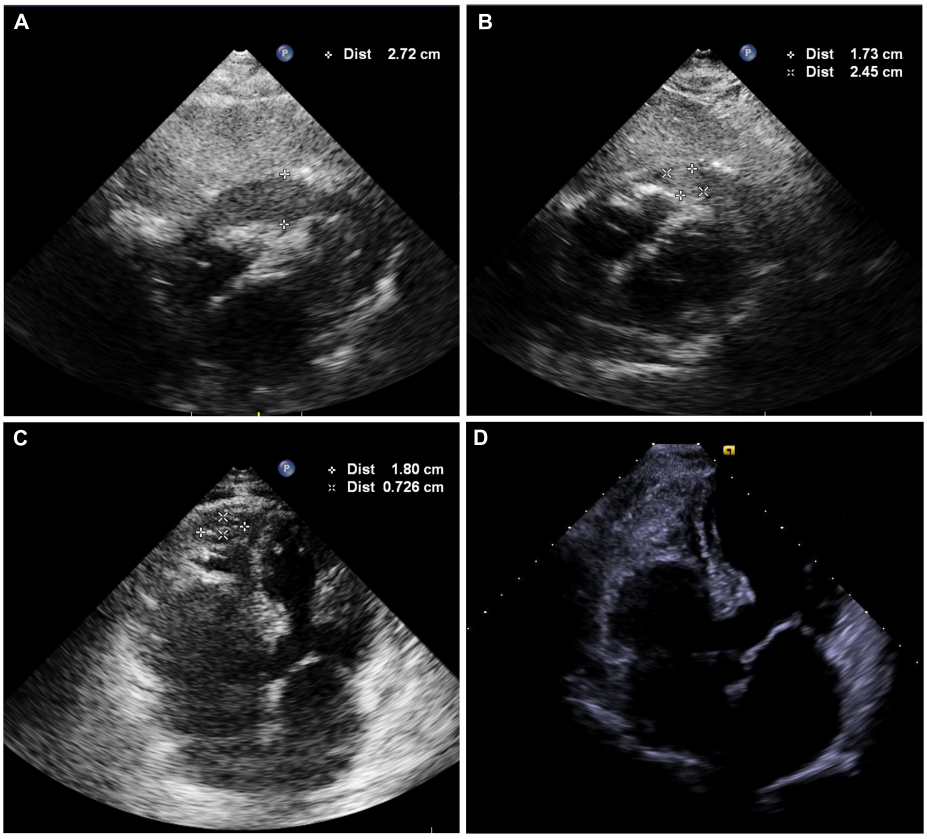

FIGURE 3
(A) Echocardiography showing hypoechoic (rather than anechoic) areas in the pericardial space. **(B)** Echocardiography shows mild effusions with a round hyperechoic thrombus in the vicinity of the right ventricular apex after intrapericardial thrombus aspiration. **(C)** Image showing shrinkage of the round thrombus, which appears as a strip in the vicinity of the right ventricular apex on the second postoperative day (**Supplementary Video 8**). **(D)** Echocardiography was obtained 2 weeks after discharge and showed no thrombus or pericardial effusion (**Supplementary Video 9**).

thrombosis was detected during the early stage in this patient. Emergency surgery is inevitable in patients with complete pericardial thrombosis or hemodynamic instability. We do not recommend the routine use of intrapericardial thrombus aspiration utilizing a thrombus aspiration catheter for the management of pericardial thrombosis; however, in selected cases, it may serve as a potential percutaneous method that obviates the need for open chest surgery and the ensuing morbidity. Whether the closure device should be deployed to seal the site of leakage for tears located within the LAA remains questionable. Our single-center experience supports this operation; however, few studies have discussed this topic, and future investigations are warranted to validate our findings.

Conclusion

Pericardial thrombosis is a life-threatening emergency, and pericardiocentesis for management of this complication is often challenging; therefore, surgical treatment remains the only definitive therapeutic strategy. In this report, we highlight the effectiveness of a percutaneous technique of intrapericardial thrombus aspiration using a dedicated thrombus aspiration catheter in selected patients with early pericardial thrombosis.

Data availability statement

The original contributions presented in this study are included in the article/**Supplementary material**, further inquiries can be directed to the corresponding authors.

Ethics statement

The studies involving human participants were reviewed and approved by the Ethics Committee of Xinhua Hospital Affiliated to Shanghai Jiao Tong University School of Medicine.

The patients/participants provided their written informed consent to participate in this study.

Author contributions

B-FM and C-QW wrote the manuscript. Q-SW and Y-GL revised the manuscript and were in charge of the design of this study. All authors approved the manuscript for publication.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Supplementary material

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References

1. Harnek J, James S, Lagerqvist B. Coronary artery perforation and tamponade- incidence, risk factors, predictors and outcomes from 12 years' data of the SCAAR registry. *Circ J.* (2019) 84:43–53. doi: 10.1253/circj.CJ-19-0757

2. Munir MB, Khan MZ, Darden D, Pasupula DK, Balla S, Han FT, et al. Pericardial effusion requiring intervention in patients undergoing percutaneous left atrial appendage occlusion: prevalence, predictors, and associated in-hospital adverse events from 17,700 procedures in the United States. *Heart Rhythm.* (2021) 18:1508–15. doi: 10.1016/j.hrthm.2021.05.017

3. Rezq A, Basavarajaiah S, Latib A, Takagi K, Hasegawa T, Figini F, et al. Incidence, management, and outcomes of cardiac tamponade during transcatheter aortic valve implantation: a single-center study. *JACC Cardiovasc Interv.* (2012) 5:1264–72. doi: 10.1016/j.jcin.2012.08.012

4. Spodick DH. Acute cardiac tamponade. *N Engl J Med.* (2003) 349:684–90. doi: 10.1056/NEJMra022643

5. Luis SA, Kane GC, Luis CR, Oh JK, Sinak LJ. Overview of optimal techniques for pericardiocentesis in contemporary practice. *Curr Cardiol Rep.* (2020) 22:60. doi: 10.1007/s11886-020-01324-y

6. Reddy VY, Sievert H, Halperin J, Doshi SK, Buchbinder M, Neuzil P, et al. Percutaneous left atrial appendage closure vs warfarin for atrial fibrillation: a randomized clinical trial. *JAMA.* (2014) 312:1988–98. doi: 10.1001/jama.2014.15192

7. Holmes DR Jr, Kar S, Price MJ, Whisenant B, Sievert H, Doshi SK, et al. Prospective randomized evaluation of the watchman left atrial appendage closure device in patients with atrial fibrillation versus long-term warfarin therapy: the PREVAIL trial. *J Am Coll Cardiol.* (2014) 64:1–12. doi: 10.1016/j.jacc.2014.04.029

8. Boersma LV, Schmidt B, Betts TR, Sievert H, Tamburino C, Teiger E, et al. Implant success and safety of left atrial appendage closure with the WATCHMAN device: peri-procedural outcomes from the EWOLUTION registry. *Eur Heart J.* (2016) 37:2465–74. doi: 10.1093/eurheartj/ehv730

9. Tzikas A, Shakir S, Gafoor S, Omran H, Berti S, Santoro G, et al. Left atrial appendage occlusion for stroke prevention in atrial fibrillation: multicentre experience with the AMPLATZER Cardiac Plug. *EuroIntervention.* (2016) 11:1170–9. doi: 10.4244/eijy15m01_06

10. Adler Y, Charron P, Imazio M, Badano L, Barón-Esquivias G, Bogaert J, et al. 2015 ESC Guidelines for the diagnosis and management of pericardial diseases: the task force for the diagnosis and management of pericardial diseases of the European Society of Cardiology (ESC)Endorsed by: the European Association for Cardio-Thoracic Surgery (EACTS). *Eur Heart J.* (2015) 36:2921–64. doi: 10.1093/eurheartj/ehv318

11. Cabrera JA, Saremi F, Sánchez-Quintana D. Left atrial appendage: anatomy and imaging landmarks pertinent to percutaneous transcatheter occlusion. *Heart.* (2014) 100:1636–50. doi: 10.1136/heartjnl-2013-304464

12. Reddy VY, Gibson DN, Kar S, O'Neill W, Doshi SK, Horton RP, et al. Post-approval U.S. experience with left atrial appendage closure for stroke prevention in atrial fibrillation. *J Am Coll Cardiol.* (2017) 69:253–61. doi: 10.1016/j.jacc.2016.10.010

13. Wu SJ, Fan YF, Chien CY. Surgical strategies for cardiac perforation after catheter ablation or electrophysiology study. *Int Heart J.* (2021) 62:1257–64. doi: 10.1536/ihj.21-201

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Case report: Transcatheter aortic valve replacement in a large bicuspid anatomy using the XL-Myval 32 mm

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Transcatheter aortic valve replacement (TAVR) is a recommended intervention for selected population with severe aortic stenosis (AS). Bicuspid aortic valve (BAV) anatomy has been categorized as an unfavorable anatomy for TAVR due to multiple considerations as exclusion from randomized trials in addition to the challenging and unpredictable anatomy. The anatomical constraints of BAV include the large anatomy of the annulus, sinus of Valsalva, and aorta (aortopathy), in addition to significant calcifications of the device landing zone. Most commercial transcatheter heart valves (THV) have upper dimension limits of the annulus and area in which the device can be implanted safely without significant oversizing. Myval-XL THVs (Meril Life Sciences Pvt. Ltd., India) are balloon-expandable valves (BEV) that have been developed with two new sizes, 30.5 and 32 mm, aiming to treat patients with large annulus dimensions and that exceed the upper limit of an ordinary device's sizing matrix. This case series report describes TAVR using the XL-Myval 32 mm THV in three European patients with symptomatic severe bicuspid aortic stenosis with significant calcifications and large annular dimensions exceeding the limits of the other THVs.

KEYWORDS

bicuspid aortic valve, large annulus, transcatheter aortic valve replacement, Myval-XL, BAV

Abbreviations: AS, Aortic Stenosis; AV, Aortic Valve; BAV, Bicuspid Aortic Valve; CCTA, Cardiac Computed Tomography Angiography; EOA, Effective Orifice Area; PVL, Paravalvular leakage; TAVR, Transcatheter Aortic Valve Replacement; TTE, Transthoracic Echocardiography.

Introduction

Transcatheter aortic valve replacement (TAVR) is a recommended intervention mode for patients with severe aortic stenosis (AS) with a tricuspid AV anatomy (1, 2). TAVR for AS and bicuspid aortic valve (BAV) disease remains challenging due to multiple anatomical and technical obstacles. BAV is considered an unfavorable anatomy for TAVR due to its exclusion from randomized trials. In addition, there is no consensus about the effectiveness of TAVR in BAV in comparison to surgical aortic valve replacement. The available evidence of TAVR in BAV has been derived from observational registries and experts' opinions (3). A critical challenge within BAV is the relatively large aortic root anatomy, including annulus diameters, that might exceed the sizing matrix of the available commercial TAVR devices (4), in addition to the associated high calcium burden, fused raphe, and aortic dilatation. Here we present a case series of three patients with low surgical risk and severe calcific BAV stenosis with large annular dimensions, treated with the novel balloon-expandable (BE) transcatheter Myval-XL 32 mm valve. All procedural characteristics, in addition to 30-day clinical and hemodynamic outcomes, are reported to describe the performance of the large new device in such a challenging anatomy.

Patient 1

The first patient was a 76-year-old man with severe symptomatic AS. A transthoracic echocardiogram (TTE) showed a transvalvular velocity of 5.1 m/s, with maximum and mean pressure gradients (PG) of 100 and 60 mmHg, respectively. The AV effective orifice area (EOA) was 0.6 cm^2 with mild aortic regurgitation and a reduced left ventricular (LV) ejection fraction (EF) of 35%. A preprocedural-cardiac computed tomography angiography (CCTA) confirmed a severely calcified BAV, Sievers' type 1-a, with a calcified raphe and annular calcific nodule (5 × 5 mm). The measurements showed a very large annulus with a perimeter of 99 mm, a perimeter-derived diameter of 31 mm and an area of 747 mm^2, wide sinuses (36 × 40 × 44 mm), inter commissural distance (ICD) 4–5 mm above the annular level of 35 mm, a sino-tubular junction diameter of 34 mm, and a mildly dilated ascending aorta of 43 mm. The left and right coronary heights were 15 and 25 mm, respectively (**Figure 1**). After a heart-team discussion, transfemoral (TF) TAVR using the BE Myval-XL 32 mm, was decided upon. The baseline ECG showed a normal sinus rhythm, a PR interval of 172 ms, and a QRS duration of 110 ms. The TVH was introduced through the right common femoral artery (CFA) after local anesthesia using a 14 Fr Python sheath (Meril Life Sciences Pvt. Ltd., India). Balloon pre-dilatation was performed using a Sapien-25 mm balloon (Edwards Lifesciences, LLC, CA, USA). The device was crimped over the Navigator delivery system (Meril Life Sciences Pvt. Ltd., India). The deployment within the annulus was performed in the tri-coplanar view (LAO15/CRA3), guided by the dense and light marking bands of the crimped device, a feature characteristic of the Myval (**Figure 1E** and **Supplementary Video 1**), with temporary pacing over the LV guidewire.

Post-implantation aortography and invasive transvalvular gradient were acceptable with Sellers' grade 1 and 5 mmHg. No intraprocedural complications were reported, and the post-procedural ECG showed no new conduction disturbance (**Figures 1G,H**). After 2 days, the patient was discharged from the hospital on Aspirin as an antithrombotic treatment. Thirty-day follow-up documented a good clinical status, with a mean trans-prosthetic PG of 7 mmHg, an AV-EOA of 2.0 cm^2 with trace paravalvular leakage (PVL), and a significantly improved EF to 50% (**Table 1**).

Patient 2

The second patient was a 79-year-old man with severe AS. A TTE revealed a maximum and mean PG of 71 and 54 mmHg, respectively, an EOA of 0.7 cm^2, and a preserved LV-EF. A preprocedural-CCTA showed a calcified BAV Sievers type 1-a, with a large annular calcific nodule (10 × 15 mm). The measurements documented a large annulus with a perimeter of 101 mm, a perimeter-derived diameter of 31.5 mm, an area of 774 mm^2, an ICD of 35 mm, and wide sinuses (**Figure 2**). A 32 mm Myval-XL was selected for TF-TAVR with the risk of annular injury due to the annular calcium chunk (not extending to the LVOT). Balloon pre-dilatation was performed using a Sapien-25 mm balloon. The device deployment was performed using the Navigator delivery system in the tri-coplanar view (RAO4/CRA5) with pacing over the LV guidewire. Final aortography (**Supplementary Video 2**) and invasive transvalvular gradient revealed Sellers' grade 0 and 5 mmHg. The post-procedural ECG showed no alteration of the conduction (**Figures 2F,G**). The patient had an uneventful in-hospital course and was discharged on day 2 with antiplatelet monotherapy (Aspirin). The patient's 30-day outcomes were uneventful (**Table 1**).

Patient 3

The third patient was a 69-year-old man with symptomatic severe AS and a history of paroxysmal atrial fibrillation and coronary intervention. A TTE confirmed severe AS with an EOA of 0.8 cm^2 and preserved LV-EF. A preprocedural-CCTA disclosed a severely calcified BAV Sievers type 1-a, with calcified raphe and an Agatston calcium score of 6132 and calcium volume of 4757 mm^3 from a non-contrast CT-scan (**Figure 3** and **Supplementary Video 3**). The measurements revealed a

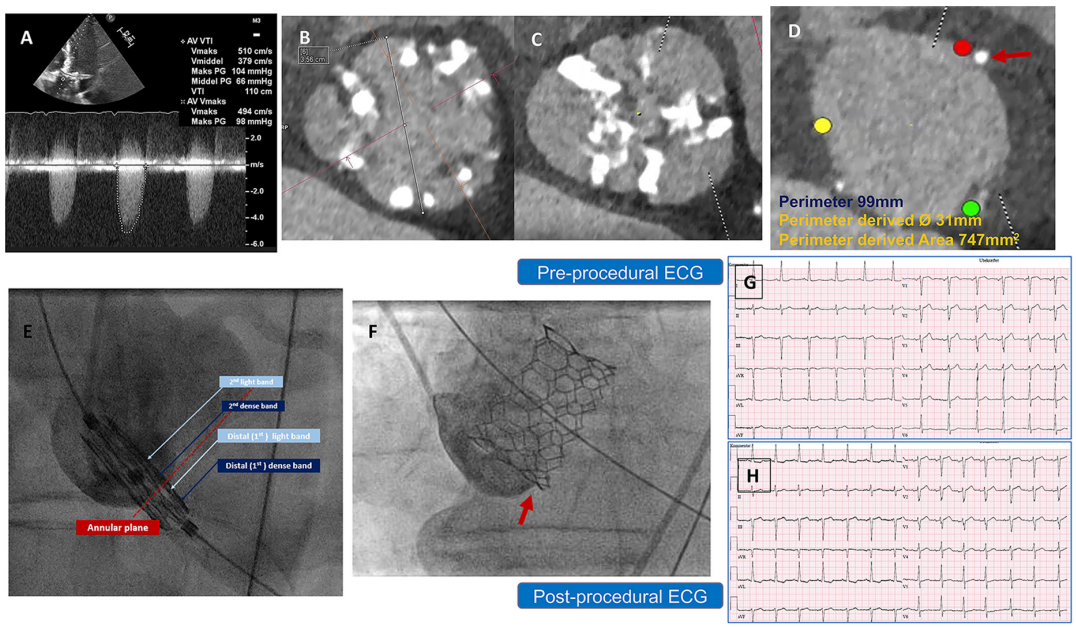

FIGURE 1
(A) Preprocedural AV CWD recording from Apical 5 chamber view, with high maximum velocity = 5.1 m/s, mean PG = 66 mmHg **(B)** inter commissural distance (4–5 mm) above the annular level = 35 mm, **(C)** AV significant calcification at the raphe level **(D)** annulus measurement showing small annular calcific nodule, and **(E)** illustration of the Myval design showing the dense (dark blue arrows), light bands (light blue arrows) and deployment within the annular plane (red dotted line) to obtain a proper position and implantation depth. **(F)** Implantation depth **(G,H)** pre and postprocedural ECG without changes. AV, Aortic valve, BAV, Bicuspid Aortic Valve, CCTA, Cardiac Computed Tomography, CWD, continuous wave Doppler, ECG, Electrocardiogram, PG, Pressure Gradient.

large annulus with a perimeter-derived diameter of 32.7 mm, a perimeter of 102.6 mm, a perimeter-derived area of 812 mm^2, an ICD of 36 mm, wide sinuses (41 × 43 × 44 mm), and an ascending aorta of 42 mm. The left and right coronary heights were 12 and 19 mm, respectively. The decision was made to implant a 32 mm Myval-XL. A baseline ECG showed a first-degree AV block (PR interval of 222 ms) and a QRS of 104 ms. The patient was operated on through the right CFA. A Sentinel Cerebral Protection System (Boston Scientific, Marlborough, MA, USA) was inserted successfully from the right radial artery. A transvenous pacing lead through the left femoral vein was used for temporary pacing. Balloon pre-dilatation was performed using a True Dilation® balloon-26 mm (Bard Peripheral Vascular, Temple, AZ) (**Supplementary Video 4**). The device deployment was performed using the standard delivery system (**Supplementary Video 5**). The final aortography revealed Sellers' grade of 0 (**Supplementary Video 6**). The patient was monitored in the hospital for 7 days for any new conduction disturbance requiring permanent pacemaker implantation. Fortunately, no changes were noticed in the pre-discharge ECG. Afterward, the patient was discharged in good condition with Dabigatran 150 mg twice daily as an antithrombotic. The patient's 30-day follow-up confirmed good clinical and hemodynamic outcomes (**Table 1** and **Supplementary Videos 7–9**).

TABLE 1 Thirty-day follow-up for each of the three patients.

	Patient 1	Patient 2	Patient 3
Pacemaker implantation	No	No	No
Renal impairment	No	No	No
Body surface area, m^2*	2.18	2.31	2.39
Body mass index, kg/m^2	35.1	32.8	30.4
TTE-follow up			
Trans prosthetic Mean PG, mmHg	7	4	7
EOA, cm^2	2.0	†	2.7
EOA index	0.92‡		1.1
PVL	Trace	Trace	Trace
LV-EF%	50	50	55

*The three patients have relatively large BSA and high BMI. †EOA was not measured due to the missing mandatory parameters for the continuity equation. ‡EOA index is acceptable regarding the patients' high BMI. TTE, Trans thoracic echocardiogram; PG, Pressure gradient; EOA, Effective orifice area; PVL, paravalvular leakage; LV-EF, Left ventricle-ejection fraction.

Discussion

With the extension of TAVR indications into lower surgical risk and younger populations, more BAV is expected to be

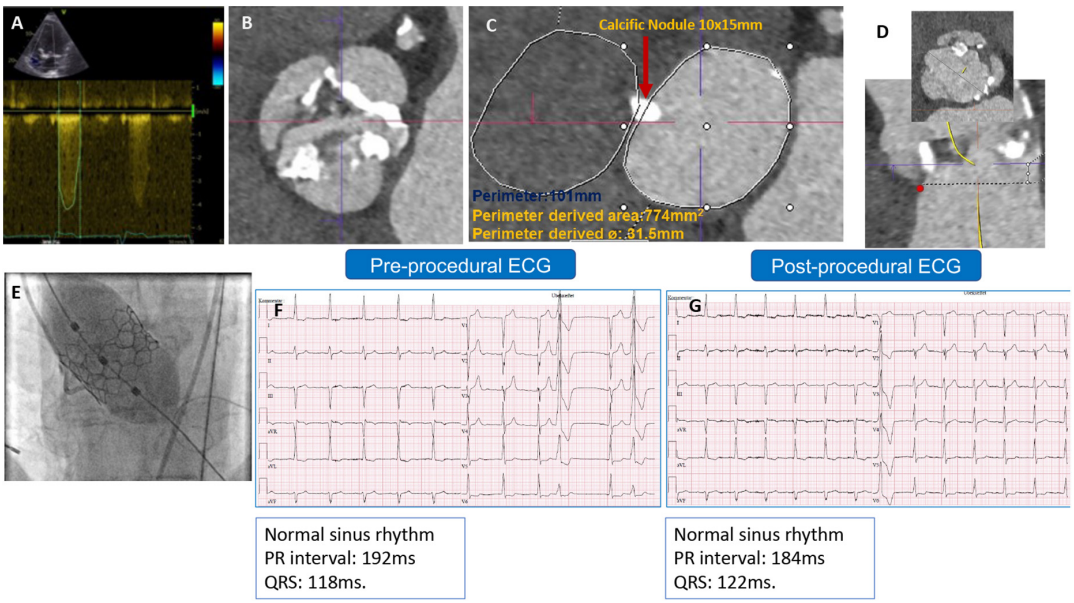

FIGURE 2
(A) Preprocedural AV continuous wave Doppler with high maximum velocity, **(B)** preprocedural MSCT showing BAV with partially fused L-R raphe and significant AV calcifications, **(C)** annulus measurement showing large annulus with large annular calcific nodule, **(D)** LVOT profile view (no calcification) and ICD 4–5 mm above the annulus = 35 mm, **(E)** Myval-XL 32 mm deployment, **(F)** preprocedural ECG, and **(G)** postprocedural ECG without changes.

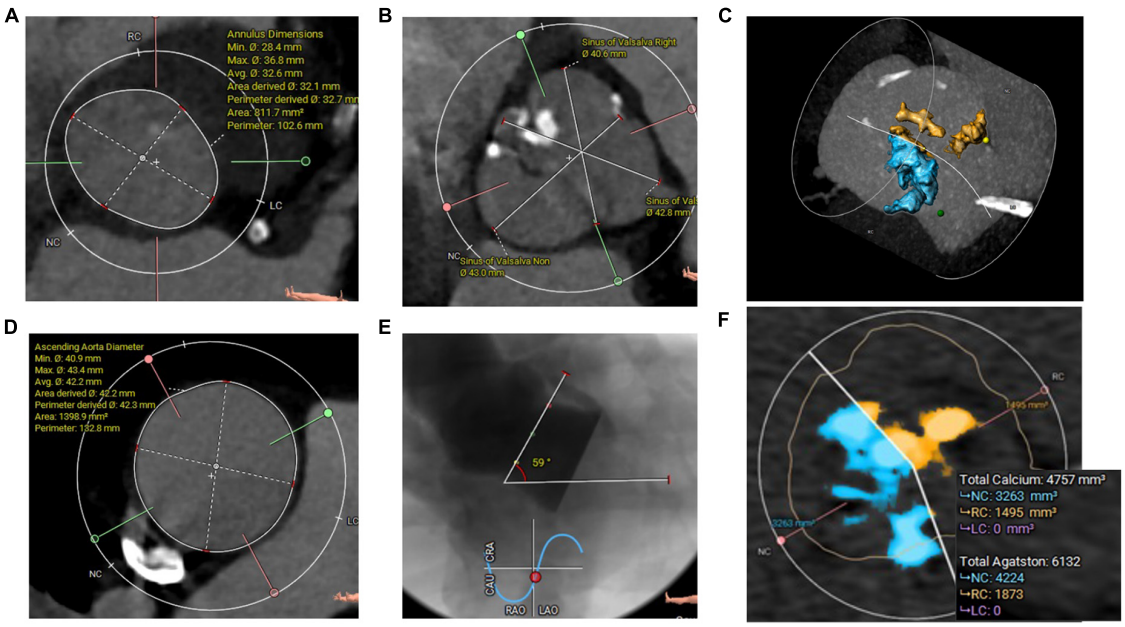

FIGURE 3
(A) CCTA Annulus measurements with a very large area, **(B)** wide sinuses, **(C)** all calcifications (3D Hockey Puck-MIP view), **(D)** ascending aorta diameters, **(E)** angiographic aortic angulation showed horizontal aorta, and **(F)** aortic valve calcifications (Agatston and volume mm^3) measured from non-contrast CT scan.

treated. BAV disease is associated with multiple anatomical constraints such as extensive calcifications and large anatomy (4, 5), so surgical intervention is still favored for those populations (1, 6). This case report series describes the successful implantation of a 32 mm Myval-XL and the 30-day outcome for three European patients with severe

BAV stenosis and large annulus dimensions exceeding the limits of all available commercial balloon- or self-expandable transcatheter heart valves (THVs). The Myval THV received a CE mark on June 2019 and introduced five extra sizes, three intermediate (21.5, 24.5, and 27.5 mm) and two extra-large (XL) sizes (30.5 and 32 mm). The included patients are relatively young with low surgical risk, so they may become exposed to future device re-intervention (Valve-in-Valve procedure) and may benefit from a rather large second device and a lower risk of PPM. The CCTA analysis has revealed high-risk features, such as significant calcifications within the raphe and the annulus, with the risk of annular rupture, especially with BEV. Hence, the SEV is frequently selected with such features and BAV. For the three above cases, the ICD diameter was larger than the annular diameter, which revealed a flared configuration of the BAVs device landing zone.

Because the maximum limit of the native annulus area and perimeter-derived diameter for other BEVs are 683 mm^2 and 29.5 mm, and for SEV, the maximum annulus perimeter is 94.2 mm, and the maximum perimeter-derived diameter is 30 mm, a 32 mm Myval-XL was the only THV fitting with this anatomy without the need for significant oversizing, which has been performed previously with such large anatomy (7, 8). From a technical perspective, significant oversizing might carry the risk of substantial deformation of the leaflet and stent geometry and, subsequently, lead to early structural failure, which may be less with a device designed specifically for such anatomy.

The recently introduced Navitor Titan THV (Abbott, USA) can fit larger anatomy better than other valves (9), with the limits of a 707 mm^2 annulus area, 30 mm annulus diameter, and a perimeter of 95 mm, which is still below the range of measurements described in this report. A TAVR within large anatomy carries the risk of device instability, embolization, or the presence of significant residual PVL, especially with the significant calcifications and asymmetric valve opening of BAV, which are not encountered in the three cases in this report, emphasizing the "stable behavior" during the implantation, in addition to the acceptable post-procedural ECG changes. With the use of the 14-Fr-Python sheath, which accommodates the device through the $\geq$ 6.5 mm CFA diameter, only two Perclose-ProGlide (Abbott, USA) were used to successfully close the CFA without residual bleeding in the three patients. The mean procedural time and contrast amounts were 36 min and 70 ml, respectively. These findings are consistent with the initial report on Myval's safety and efficacy in BAV (10).

Conclusion

The implantation of the 32 mm Myval-XL device in a large annulus and BAV appears safe with promising and acceptable hemodynamic and clinical outcomes. This might be considered a step toward further expansion of the clinical indication of the TAVR practice and highlights the concept of patient-specific device selection by offering a dedicated device specifically designed for patients with large anatomy.

Data availability statement

The original contributions presented in this study are included in the article/**Supplementary material**, further inquiries can be directed to the corresponding authors.

Ethics statement

Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

Author contributions

AE, OS, CT, and HN: conceptualization of the report and writing and drafting of the manuscript. CT, JE-G, TT, and HN: investigations. OS, CT, and HN: supervision. PS, MA, HE, JE-G, and TT: manuscript review and editing. All authors contributed to the article and approved the submitted version.

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Conflict of interest

Author AE received a scholarship from the University of Galway. Author CT received a proctor fee from Meril Life, Edwards, Boston, and a lecture fee from Terumo and Bristol-Myers Squibb.

The remaining authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fcvm.2022.1045280/full#supplementary-material

References

1. Vahanian A, Beyersdorf F, Praz F, Milojevic M, Baldus S, Bauersachs J, et al. 2021 ESC/EACTS Guidelines for the management of valvular heart disease. *Eur Heart J.* (2021) 43:561–632. doi: 10.1093/ejcts/ezac209

2. Otto CM, Nishimura RA, Bonow RO, Carabello BA, Erwin JP 3RD, Gentile F, et al. 2020 ACC/AHA Guideline for the management of patients with valvular heart disease: A report of the american college of cardiology/american heart association joint committee on clinical practice guidelines. *J Am Coll Cardiol.* (2021) 77:e25–197. doi: 10.1161/CIR.0000000000000923

3. Vincent F, Ternacle J, Denimal T, Shen M, Redfors B, Delhaye C, et al. Transcatheter aortic valve replacement in bicuspid aortic valve stenosis. *Circulation.* (2021) 143:1043–61. doi: 10.1161/CIRCULATIONAHA.120.048048

4. Xiong, TY, Ali WB, Feng Y, Hayashida K, Jilaihawi H, Latib A, et al. Transcatheter aortic valve implantation in patients with bicuspid valve morphology: a roadmap towards standardization. *Nat Rev Cardiol.* (2022) 1–16. doi: 10.1038/s41569-022-00734-5 [Epub ahead of print].

5. Michelena HI, Della Corte A, Evangelista A, Maleszewski JJ, Edwards WD, Roman MJ, et al. International consensus statement on nomenclature and classification of the congenital bicuspid aortic valve and its aortopathy, for clinical, surgical, interventional and research purposes. *Ann Thorac Surg.* (2021) 112:e203–35. doi: 10.1016/j.athoracsur.2020.08.119

6. Varc-3-Committee, Genereux P, Piazza N, Alu MC, Nazif T, Hahn RT, et al. Valve Academic Research Consortium 3: updated endpoint definitions for aortic valve clinical research. *Eur Heart J.* (2021) 42:1825–57. doi: 10.1093/eurheartj/ehaa799

7. Barr P, Ormiston J, Stewart J, Nand P, Ramanathan T, Webster M. Transcatheter aortic valve implantation in patients with a large aortic annulus. *Heart Lung Circ.* (2018) 27:e11–4. doi: 10.1016/j.hlc.2017.08.025

8. Buchanan KD, Rogers T, Shults C, Ben-Dor I, Satler LF, Waksman R. Successful transcatheter aortic valve replacement in an oversized 800mm(2) annulus and bicuspid aortic valve. *Cardiovasc Revasc Med.* (2018) 19:65–7. doi: 10.1016/j.carrev.2018.03.023

9. Bajoras V, Nuyens P, Vanhaverbeke M, Wang X, Bieliauskas G, De Backer O, et al. TAVR with the novel Navitor Titan transcatheter heart valve to treat aortic stenosis patients with large aortic annuli. *Cardiovasc Revasc Med.* (2022) 40S:120–2. doi: 10.1016/j.carrev.2022.03.028

10. Elkoumy A, Jose J, Terkelsen CJ, Nissen H, Gunasekaran S, Abdelshafy M, et al. Safety and efficacy of myval implantation in patients with severe bicuspid aortic valve stenosis—a multicenter real-world experience. *J Clin Med.* (2022) 11:443. doi: 10.3390/jcm11020443

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Case report: Percoronary device closure of tortuous coronary artery fistula into left atrium

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Surgical ligation and transcatheter occlusion are the mainstream for the treatment of coronary artery fistulas (CAFs). However, these techniques applied to tortuous and aneurysmal CAF, especially those draining into left-heart, have their known drawbacks. We report, a successful percoronary device closure of such CAF, originating from left main coronary artery and draining into left atrium, through a left subaxillary minithoracotomy. Through a puncture on the distal straight course, we occluded CAF exclusively under transesophageal echocardiography guidance. Complete occlusion was achieved. It's a simple, safe, and effective alternative for tortuous, large, and aneurysmal CAFs draining into the left heart.

KEYWORDS

coronary artery fistula, percoronary, device closure, minithoracotomy, occlusion

Introduction

Coronary artery fistula (CAF) is a rare congenital heart disease. Surgical ligation and transcatheter device closure (TCC) are the mainstream for treatment of CAF (1–3). However, surgical ligation is associated with significant trauma, morbidity, discomfort, an unsightly scar and the use of cardiopulmonary bypass. The TCC is a preferable way of treatment, but is troublesome in highly tortuous, large, and aneurysmal CAF, especially those draining into the left heart chamber. We present an alternative and achievable technique of percoronary device closure (PDC) of CAF, which was tortuous, large, aneurysmal and draining into the left atrium (LA), under exclusive transesophageal echocardiography (TEE) guidance.

Case presentation

A 32-year-old man was admitted with a left scapular pain and diagnosed with a CAF by TEE and computed tomographic angiography (CTA). The TEE showed the CAF originated from main coronary artery (MCA) and drained into the LA with a continuous left-to-left shunt. The maximum inner diameter of the CAF was 11 mm and the left ventricular end diastolic diameter was 59 mm. The CTA was performed to delineate the anatomy of the CAF (**Figures 1A–C**). A continuous murmur was detected in the parasternal left 2nd and 3rd intercostal space. Non-specific electrocardiogram abnormalities were found. A bone-reserved thoracic CTA was also performed before the procedure to determine the exact location of the incision and procedure planning (**Figure 1D**). The informed consent was obtained.

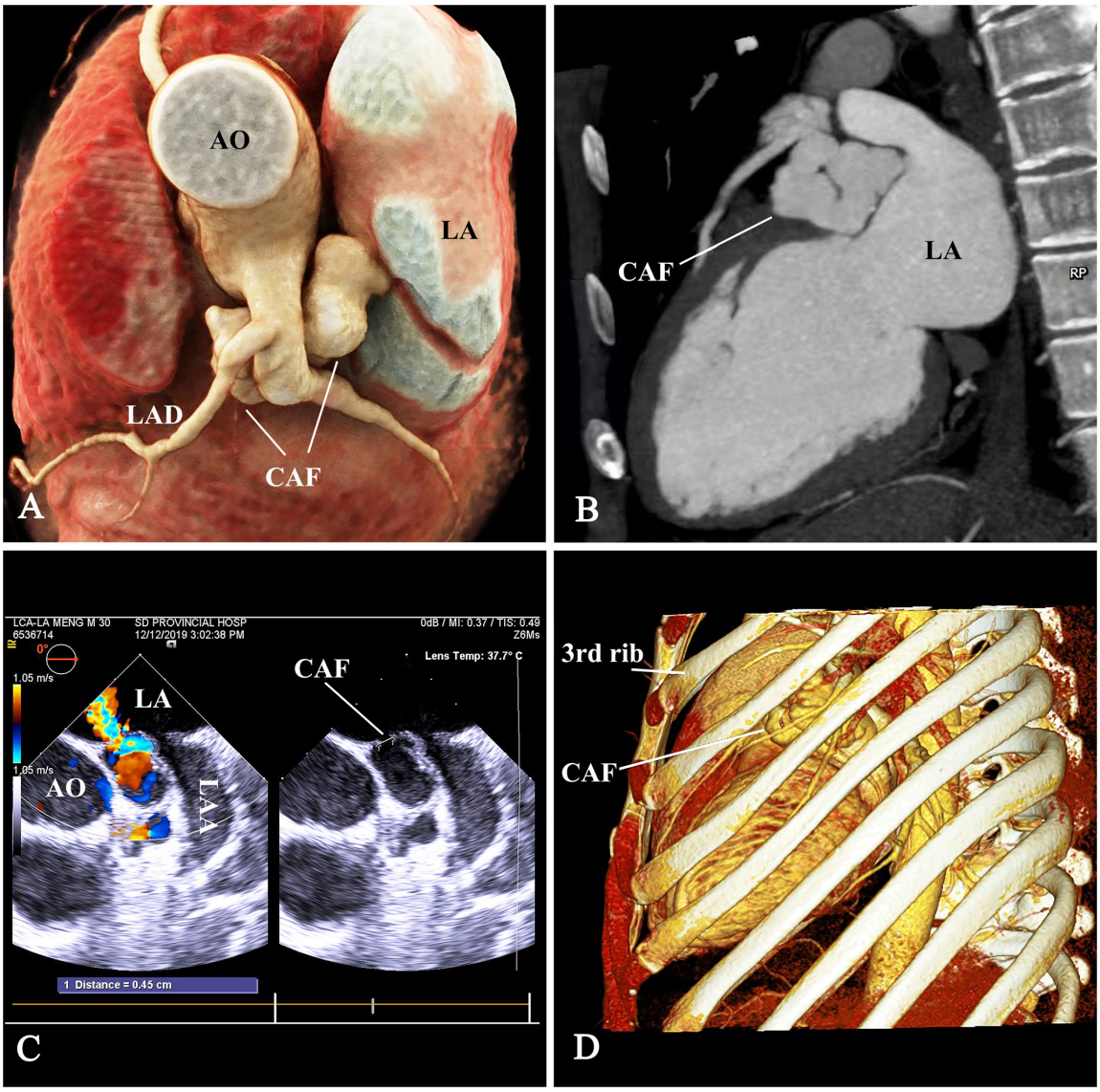

FIGURE 1
The anatomy of CAF. **(A,B)** The tortuous aneurysmal CAF originated from main coronary artery (MCA), made a 180° turn underneath the LAD and MCA, and coursed backward to drain into the LA. **(C)** The drainage opening measuring 4.5 mm, was located at the LA roof between the aortic root and LAA. **(D)** The thoracic computed-tomographic-angiography to determine the access. CAF, coronary artery fistula; AO, ascending aorta; LAD, left anterior descending branch; LA, left atrium; LAA, left atrial appendage.

Under general anesthesia, the patient was placed in a right lateral position. A 7-cm subaxillary incision was made in the left 4th intercostal space. The pericardium was incised and cradled. An ordinary (size 8 mm) ventricular septal occluder (Starway Medical Technology, Inc, Beijing, China), connected with a stay-in suture (5-0 Polydioxanone, Ethicon, Somerville), was selected and retracted into the loading sheath. **Figure 2** showed the steps of percoronary device closure of CAF. A 15-min occlusion test of CAF was well-tolerated; there was no evidence of myocardial ischemia. The occluder was scrutinized by using a push-pull maneuver repeatedly and released after satisfactory assessment. The device stay-in suture was removed before the purse-string suture snugly tied. The pericardium and incision were closed in layers with a drainage tube.

Complete occlusion was achieved immediately after device release. Serial CKMB and troponin levels in the first 48 h were within normal limits. The patient recovered uneventfully and discharged 7 days postoperative. The murmur disappeared post-occlusion. Antiplatelet therapy was maintained on aspirin (100 mg/day) and clopidogrel (75 mg/day) for a year. Medical evaluations including electrocardiograms, TTE, CTA, and stress tests were reviewed during the follow-up period of 18 months. These records demonstrated an appropriate device position and complete occlusion of the CAF and no complications (**Figures 3A–D**). The **Supplementary Figure 1** shows the device and stay-in-suture use.

Comment

Coronary artery fistulas originating from the right coronary artery account for 50–60% of cases and often drain into the right heart (80%). Only 0.7% of cases originate from MCA and only 5–6% drain into the LA (1–3).

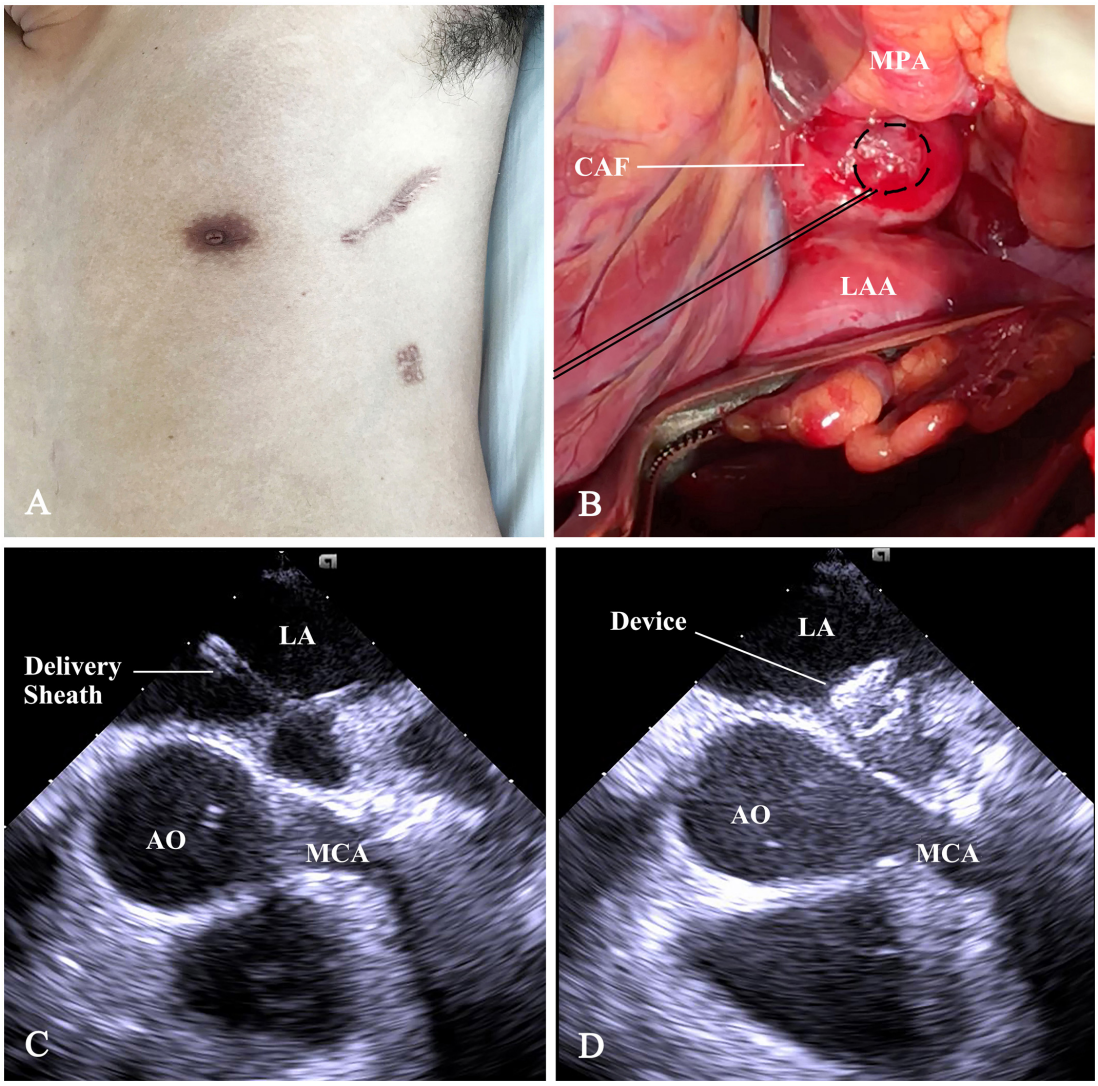

FIGURE 2
Schematic presentation of percoronary device closure of CAF. **(A)** The subaxillary incision. **(B)** The distal straight section of CAF was exposed between the main pulmonary artery (MPA), root of AO, and the LAA. A purse-string suture was placed on it. **(C)** Following a puncture in purse-string, a flexible guidewire was advanced into LA. Then, a 6F short delivery sheath was fed over the wire into the LA. **(D)** The device was deployed and positioned at the drainage orifice of the CAF. CAF, coronary artery fistula; AO, ascending aorta; LAA, left atrial appendage; LA, left atrium; MPA, main pulmonary artery; MCA, main coronary artery.

The common treatment for CAF is surgical and TCC. The choice of the technique depends on its morphology, course, tortuosity, and the presence of aneurysmal CAF.

The major advantages of TCC over surgery include the avoidance of cardiopulmonary bypass, median sternotomy and the related complications, short recovery time, lower morbidity, and improved cosmetic results. The TCC approaches are antegrade venous, retrograde arterial, or arteriovenous loop. If the drainage orifice locates in the right heart chamber and is easy to access, the antegrade venous approach is the preferred route, whereas if access is difficult, the arteriovenous loop method is a better choice (2, 3). Another access route for highly tortuous CAF, draining into the right heart, is a perventricular approach (4). However, TCC of fistulas, draining into the left heart chambers, is challenging and can only be achieved by retrograde arterial, or "arterioarterial loop" approach.

The proximal CAF arising from MCA is highly recommended to close, as this type of fistula has the risk of increased aneurismal dilatation and rupture. When the drainage site is located in the left-heart, the CAF is particularly tortuous (3, 5). It results in not only the well-known "coronary steal phenomenon," but also a left-to-left shunt that increases a volume overload to the left heart.

Although coils can be delivered with a retrograde arterial approach through very small catheters and sheaths, the tortuous aneurismal CAF prevent the guidewire and sheath's advancement and increase the risk of rupture. Additionally, the coil occlusion in large CAF might be unstable or incomplete. Mispositioning or proximal extension of the coils may result in obstruction of coronary branches and myocardial ischemia. Incomplete occlusion might raise concerns for bacterial endocarditis.

In this case, the CAF was large, aneurysmal, and tortuous. The distal segment, which was adjacent to ascending aorta, LA, MCA (**Figures 1A**, **2B**) and in deep anatomic position, was exposed through minithoracotomy. Surgical ligation is almost unthinkable because of limited anatomic space and the risk of catastrophic

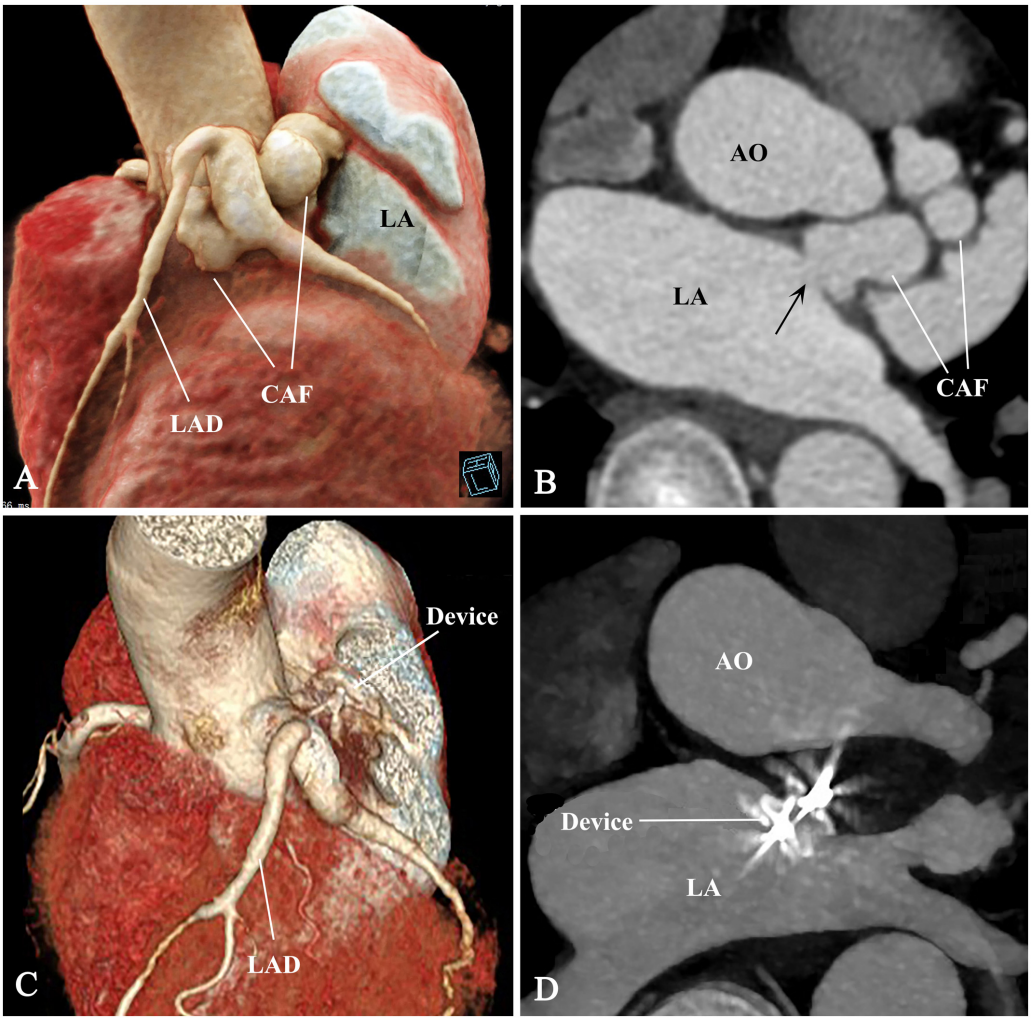

FIGURE 3
Pre- and post-procedural comparison of CAF. **(A–D)** The CAF disappeared and thrombogenesis occurred from distal to proximal segment of the CAF. CAF, coronary artery fistula; AO, ascending aorta; LAD, left anterior descending branch; LA, left atrium; arrow, fistulous drainage orifice.

bleeding. For TCC, there was not a feasible segment for positioning a double-disk device except the segment below the MCA and the drainage opening at the LA (**Figures 1A**, **3A**). Device positioning below the MCA has the risk of impingement on the MCA. Device occlusion at the fistula drainage opening should be the best choice, although the drainage site is almost inaccessible for the delivery sheath due to the acute turn and tortuous course of the CAF.

The PDC provides a straighter catheter course and a good puncture angle, avoids potential damage to the adjacent structures and femoral artery. The puncture site is chosen at the distal straight course of CAF near the drainage opening, allowing larger catheters to be used. This technique is a simple, safe, and effective alternative therapy for tortuous, large, and aneurysmal CAFs draining into the left heart chamber.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving human participants were reviewed and approved by the Ethics Committee of the First Affiliated Hospital of Shandong First Medical University. The patients/participants provided their written informed consent to participate in this study.

Author contributions

S-BS and ZK: conception, data collection, data analysis, interpretation, drafting the manuscript, and revision. ZF and LH: conception, drafting the manuscript, and critical revision. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fcvm.2023.1106420/full#supplementary-material

SUPPLEMENTARY FIGURE 1
The device can be seen attached to the delivery cable and ready to be loaded in the delivery sheath. The polydioxanone stay-in-suture can be seen passed through the center of the device under the screw, then passed back through the sheath. The inset shows the closeup of the device with stay-in-suture.

References

1. Mangukia C. Coronary artery fistula. *Ann Thorac Surg.* (2012) 93:2084–92. doi: 10.1016/j.athoracsur.2012.01.114

2. Loukas M, Germain A, Gabriel A, John A, Tubbs R, Spicer D. Coronary artery fistula: a review. *Cardiovasc Pathol.* (2015) 24:141–8. doi: 10.1016/j.carpath.2014.01.010

3. Buccheri D, Chirco P, Geraci S, Caramanno G, Cortese B. Coronary artery fistulae: anatomy, diagnosis and management strategies. *Heart Lung Circ.* (2018) 27:940–51. doi: 10.1016/j.hlc.2017.07.014

4. Zhang Q, Duan Y, Hongxin L, Wenbin G. An innovative technique of perventricular device closure of a coronary artery fistula through a left parasternal approach. *Eur Heart J.* (2017) 38:3177. doi: 10.1093/eurheartj/ehx357

5. Challoumas D, Pericleous A, Dimitrakaki I, Danelatos C, Dimitrakakis G. Coronary arteriovenous fistulae: a review. *Int J Angiol.* (2014) 23:1–10. doi: 10.1055/s-0033-1349162

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Minimally invasive closure of a progressive pseudoaneurysm of the ascending aorta: A case report

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Ascending aortic pseudoaneurysm (AAP) is rare but may cause life-threatening complications. Although the placement of a stent graft and the use of occluder devices and vascular plugs to exclude pseudoaneurysm are adopted for some patients, the management of progressive pseudoaneurysms that may rupture at any time remains a challenge that needs to be addressed. In this study, we present the case of a patient with an AAP that was caused by aortic and mitral valve replacement for the giant left ventricle. Aortic pseudoaneurysm was suspected on the basis of a spherical cystic echo (70 × 80 mm) of the ascending aorta; this pseudoaneurysm was detected by an ultrasonic cardiogram, and the diagnosis was confirmed by an aortic computed tomography angiography (CTA) examination. To prevent the unexpected rupture of a progressive pseudoaneurysm, our patient was treated with a 28- mm ASD occluder without any procedural complications. Our patient has a good prognosis, which will inspire clinicians to choose minimally invasive procedures when dealing with such high-risk cases in emergency situations.

KEYWORDS

aorta, pseudoaneurysm, minimally invasive treatment, high-risk patients, mortality

Background

Ascending aortic pseudoaneurysm (AAP) is a rare condition that occurs in less than 0.5% of all cardiothoracic surgical patients (1). If a large pseudoaneurysm is located in the retrosternal space, then there is a very high risk of massive bleeding from rupture during resternotomy (2). Redo surgery requires lengthy and meticulous tissue dissection with significantly high mortality and morbidity (3). We performed a minimally invasive closure on a patient with ascending aortic pseudoaneurysm to prevent the development of this complication.

Case presentation

A 68-year-old male was asymptomatic until a postoperative review of calf lacerations was performed. A 4/6 grade of diastolic whistling murmur in the auscultation area around the mitral valve was detected on physical examination. His history was significant for aortic valve and mitral valve replacement surgery in 2017 due to severe aortic and mitral regurgitation. A chest x-ray indicated an enlarged, tumor-like swelling in the right cardiac border and a metal shadow (**Figure 1A**). Routine blood tests revealed moderate anemia with a hemoglobin level of 77 g/L, a CRP level of 6.29 mg/L, and an albumin level of 32.50 g/L. During hospitalization, the D-dimer level was approximately 6.96 mg/L and

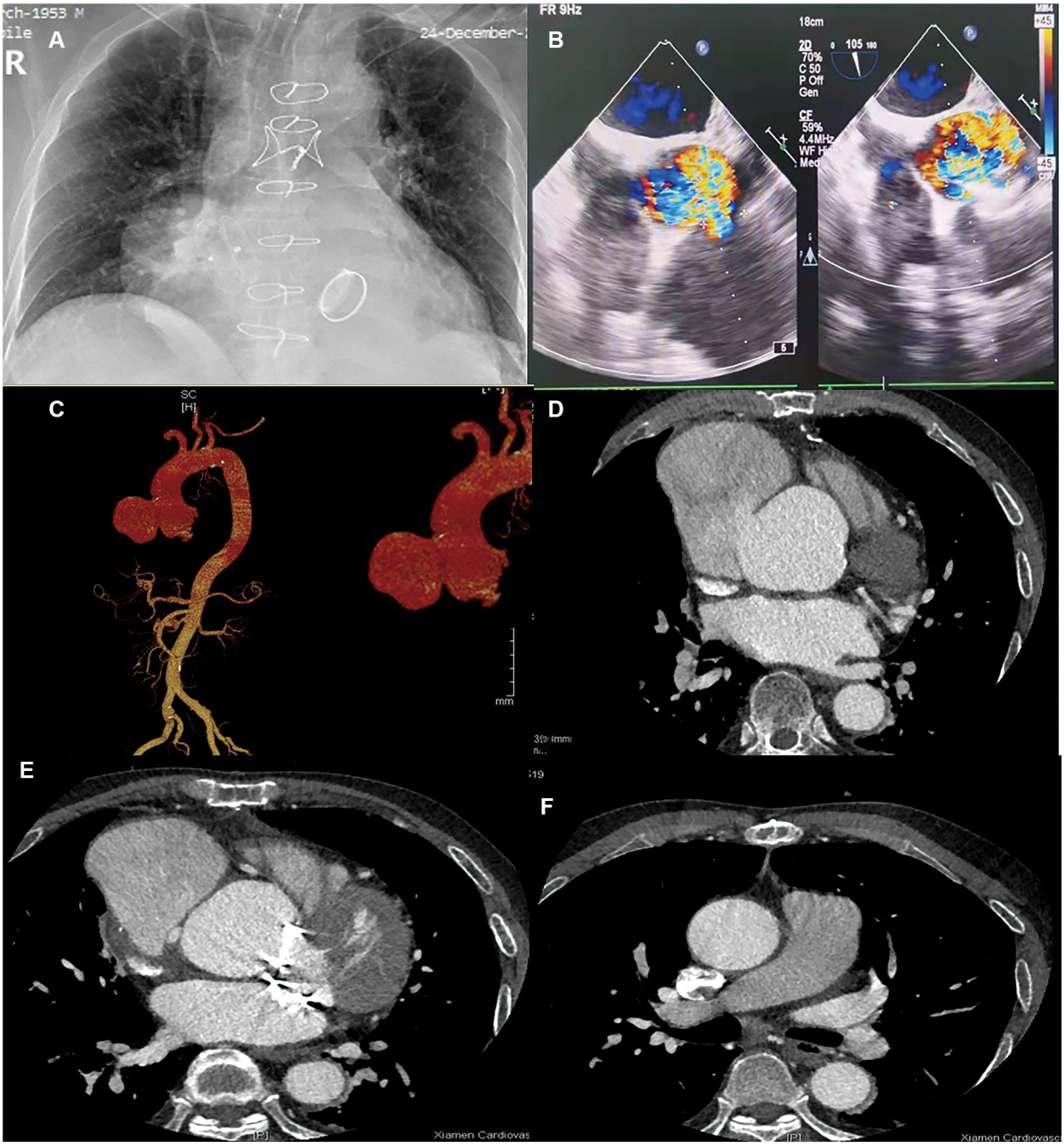

FIGURE 1
Preoperative: (A) Chest ray; (B) Cardiac ultrasound; (C–F) CT of coronary artery + thoracic and abdominal aorta [(C) Front position; (D) Tumor waist; (E) Lower border of the tumor; (F) Pulmonary bifurcation].

there was persistently elevated calcitoninogen, which indicated that a pseudoaneurysm may rupture at any time leading to sudden death. In addition, he suffered serious damage to his liver and kidney function. Other laboratory test results were normal. A cardiac ultrasound indicated that the breadth of the aortic sinus was approximately 63 mm, the sinus junction had disappeared, the aortic arch diameter was 37 mm, and the descending aortic diameter was 28 mm (Figure 1B). A CT of the coronary arteries + thoracic and abdominal aorta suggested that the ascending aorta and aortic sinus were widened, the latter to an area of 49 mm × 50 mm. The right anterior border of the ascending aorta presented with an aortic aneurysm–like structure measuring approximately 72 mm × 84 mm (Figures 1C–F). From the image, we can see that the breaking mouth from the left crown is 5.3 cm, and the breaking mouth from the right crown is 5.2 cm, with a tumor neck of 4.01 cm (Supplementary Material S1).

Compared with open surgery, minimally invasive closure greatly lowers the risk of sudden death from a punctured pseudoaneurysm. Based on the results of the inflammatory indicator and blood cultures tests, which did not show significant

bacterial infection, we utilized empirical antibiotics to manage the infection during hospitalization. After obtaining full informed consent, the patient chose minimally invasive repair of the pseudoaneurysm. After the patient was placed under general anesthesia and into the supine position, the right femoral artery was punctured with a 7F vascular sheath, and the left femoral artery was punctured with a 5F vascular sheath. The sinus duct junction was seen on the side of the greater curvature of the ascending aorta with a tumor-like deposit of contrast (**Figures 2A,B**). First, the blood pressure was lowered to

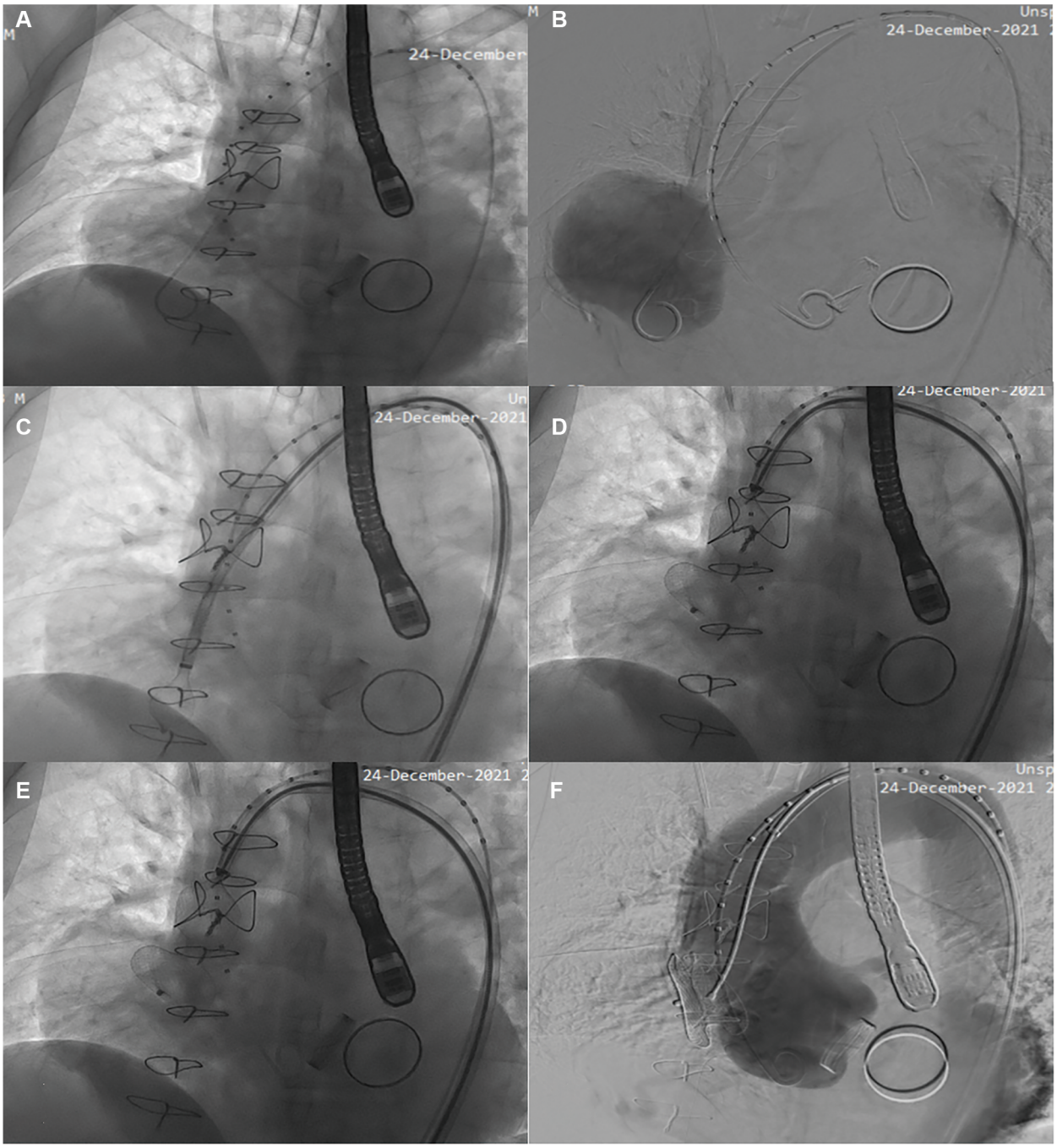

FIGURE 2
Angiography suggested that the shape of the ascending aortic sinus and the left and right coronary arteries were normal, that there was a crack at the junction of the ascending aorta, and of tumor-like deposits of a contrast agent (**A**,**B**). First, the blood pressure was reduced to approximately 70/40 mmHg. Then, a Lunderquist guidewire was fixed intra-aneurysmally, and the sheath was angled and delivered through the sheath into the atrial defect occluder. Intra-aortic angiography showed a complete sealing of the breach (**C**). After gradually increasing the pressure to 90/60 and 130/85 mmHg, the blocker was replaced and reimaged and placed in the normal position, and then the wire was rotated to release the blocker (**D**,**E**). Finally, the vehicle was observed for 5 min. A complete occlusion and good aortic valve function and occluder placement were confirmed (**F**).

approximately 70/40 mm Hg. The Lunderquist guidewire was then replaced and secured in the aneurysm. Then, the Cien-adjustable bend sheath was fed, the sheath was tilted, and the atrial defect occluder (42-28-32) was delivered through the sheath in the aneurysm. The anterior disc was opened and the fixed guidewire was gradually withdrawn posteriorly. The posterior disc was opened and the wire was delivered further forward without displacement of the occluder. Intra-aortic angiography showed a complete closure of the breach (**Figure 2C**). After gradually increasing the pressure to 90/60 and 130/85 mmHg, the blocker was placed in the normal position on separate angiograms, and the wire was then rotated to release the blocker (**Figures 2D,E**). Finally, the catheter sheath was retrieved in the descending aorta and observed for 5 min. A complete occlusion and good aortic valve function and occluder position were confirmed (**Figure 2F**). An intraoperative transesophageal ultrasound reassessment of the unaffected aortic valve function was performed (**Supplementary Material S2**).

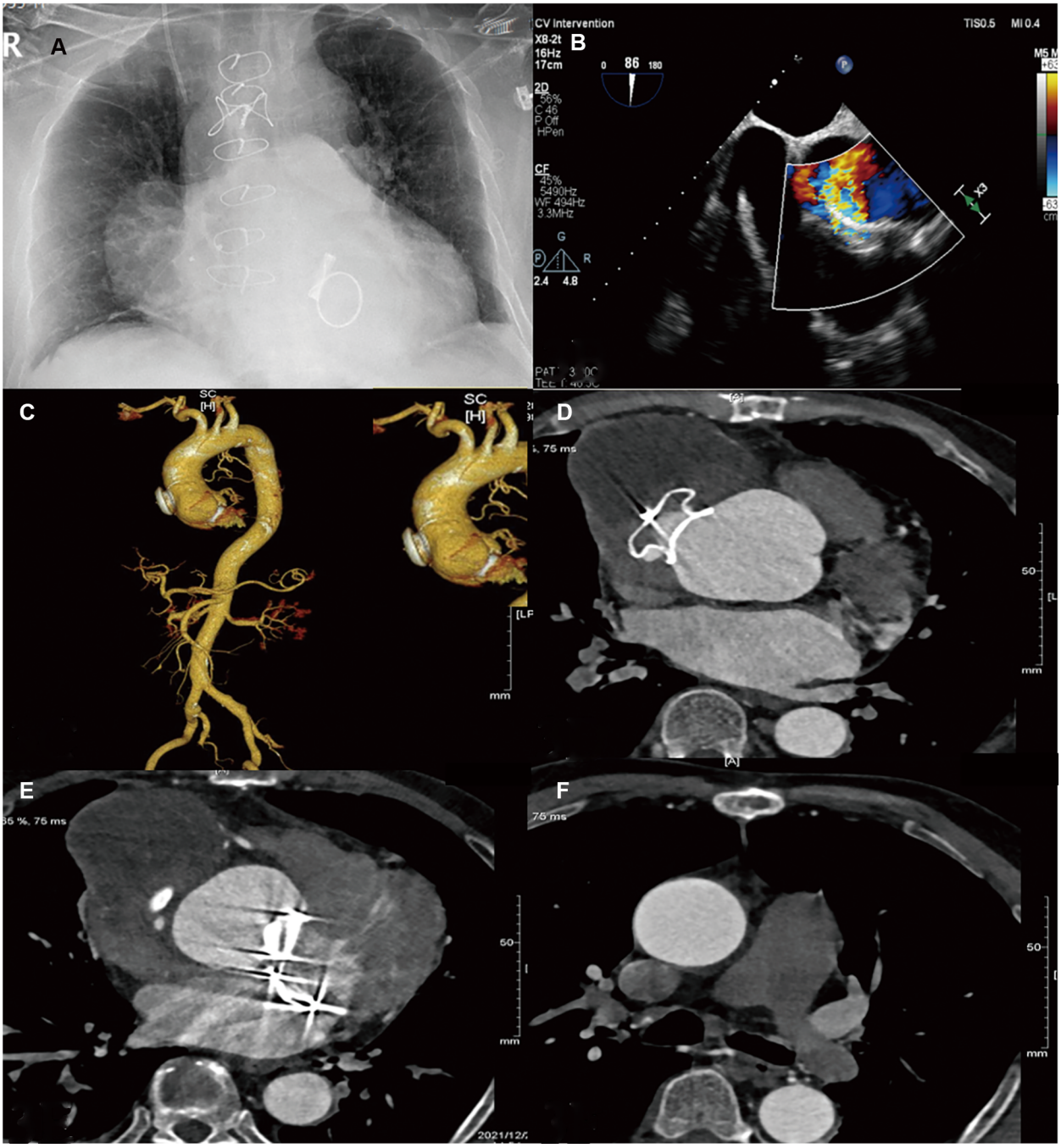

FIGURE 3
Preoperative: (A) Chest ray; (B) Cardiac ultrasound; (C–F) CT of the coronary artery + thoracic and abdominal aorta [(C) Front position; (D) Tumor waist; (E) Lower border of the tumor; (F) Pulmonary bifurcation)].

At the 3-month and 6-month follow-ups, our patient was found to have a good postoperative heart, liver, and kidney function. A postoperative chest x-ray showed no obvious abnormalities (**Figure 3A**). A postoperative echocardiography showed that the position of the AAP occluder was normal and the internal diameter of the ascending aorta had reduced (**Figure 3B**). A postoperative CT of the thoracic and abdominal aorta indicated that the occluder was in a normal position (**Figures 3C–F**).

Discussion

During cardiac surgery, an ascending aorta pseudoaneurysm can lead to serious complications. Traditionally, surgery has been the recommended approach for the correction of thoracic pseudoaneurysms (4); however, it carries a risk of 7%–41% mortality and a very bad prognosis (5). As a result, percutaneous endovascular approaches have emerged as a viable alternative to traditional therapy (6).

Some studies have reported on the adoption of techniques such as a placement of a stent graft and transcatheter aortic valve implantation and the use of occluder devices vascular plugs to exclude pseudoaneurysm (7, 8). However, little information on whether endovascular therapy is suitable for progressive pseudoaneurysm patients is available (9). In our patient, a CT of the coronary arteries + thoracic and abdominal aorta provided us with accurate anatomical information. The aneurysm was located in the noncoronary sinus with a safe distance from the right and left coronary arteries, and it did not affect the coronary blood flow, given that the AAP neck was too large for the use of coils. In addition, due to the possibility of rupture of a huge pseudoaneurysm aneurysm at any time, the current instance was not appropriate for stent graft placement. Therefore, our case can be described as one of an asymptomatic patient with an expanding AAP. To the best of our knowledge, this is the largest device ever used for postsurgical pseudoaneurysm closure. A promising sign of progress toward final AAP closure was the reduced internal diameter of the ascending aorta shown on the 3-month and 6-month imaging follow-ups.

Minimally invasive surgery avoids a reoperation of the heart and reduces the risk of poor perfusion of all organs. In this case, the patient regained his postoperative cardiac function and had significantly improved liver and kidney function. Compared with open-heart surgery, minimally invasive surgery reduces the risk of surgery and complications such as intraoperative bleeding and infection. In this case, the patient was admitted with malnutrition and moderate anemia, and the pseudoaneurysm was in the active stage of rupture. We attempted to use antibiotics along with surgery to prevent infection. Our patient was discharged from the hospital one week after surgery and showed favorable outcomes in the follow-up; therefore, this treatment may provide a new option for the emergency treatment of patients with high-risk aortic pseudoaneurysms.

In conclusion, the procedure reduced not only the risk of death but also the incidence of complications. Our case may strengthen the understanding of surgeons about minimally invasive treatment of patients with high-risk pseudoaneurysms and inspire clinicians to choose a minimally invasive approach. Eventually, the shift in treatment philosophy may reduce perioperative mortality rates.

Data availability statement

The original contributions presented in the study are included in the article/**Supplementary Material;** further inquiries can be directed to the corresponding author.

Ethics statement

Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

Author contributions

All authors contributed to the article and approved the submitted version.

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Conflict of interest

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Supplementary material

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SUPPLEMENTARY IMAGE 1
The progression of the patient's condition.

SUPPLEMENTARY IMAGE 2
The location of breaking mouth, the tumor neck was 4.01 cm.

SUPPLEMENTARY IMAGE 3
The breaking mouth from the right crown was 5.2 cm.

SUPPLEMENTARY IMAGE 4
The breaking mouth from the left crown was: 5.3 cm.

SUPPLEMENTARY VIDEO 1
An intraoperative transesophageal ultrasound reassessment of unaffected aortic valve function.

SUPPLEMENTARY VIDEO 2
Pre- and post-operative comparison of intraoperative transesophageal ultrasound of aortic valve function.

SUPPLEMENTARY TABLE 1
CARE Checklist.

References

1. Li X, Zhou H, Zhang R, Zhao J, Li T, Zhang Y, et al. Case report: ascending aortic pseudo-aneurysm following ventricular septal defect repair in a 4-year-old girl. *Front Pediatr*. (2021) 9:576527. doi: 10.3389/fped.2021.576527

2. Gadhinglajkar S, Sreedhar R. Surgery for pseudoaneurysm of the ascending aorta: role of intraoperative 2-dimensional and real-time 3-dimensional transesophageal echocardiography. *J Cardiothorac Vasc Anesth*. (2010) 24(5):906–8. doi: 10.1053/j.jvca.2009.12.011

3. Ali R, Elhosiny A, Abualnaja S, Baslaim G. Incidental finding of an aspergillus pseudoaneurysm in the ascending aorta of an immunocompetent patient. *Int Med Case Rep J*. (2021) 14:843–7. doi: 10.2147/IMCRJ.S340364

4. He C, Jiang B, Wang M, Ren P, Murtada S-I, Caulk AW, et al. mTOR inhibition prevents angiotensin II-induced aortic rupture and pseudoaneurysm but promotes dissection in apoe-deficient mice. *JCI Insight*. (2022) 7(3):e155815. doi: 10.1172/jci.insight.155815

5. Blaizot R, Desrousseaux G, Joly MP, Vie A, Lu X, Senchyna A, et al. Pseudoaneurysm following a freshwater stingray injury. *J Eur Acad Dermatol Venereol*. (2019) 33(10): e356–7. doi: 10.1111/jdv.15648

6. Kawatani Y, Kurobe H, Nakamura Y, Suda Y, Hori T. Aortic rupture due to radiation injury successfully treated with thoracic endovascular aortic repair. *J Surg Case Rep*. (2017) 2017(5):rjx092. doi: 10.1093/jscr/rjx092

7. Quevedo HC, Santiago-Trinidad R, Castellanos J, Atianzar K, Anwar A, Abi Rafeh N. Systematic review of interventions to repair ascending aortic pseudoaneurysms. *Ochsner J*. (2014) 14(4):576–85. doi: 10.3389/fcvm.2023.1134196

8. Chiocchi M, Pugliese L, D'Errico F, Tosto FD, Cerimele C, Pasqualetto M, et al. Transcatheter aortic valve implantation in patients with unruptured aortic root pseudoaneurysm: an observational study. *J Cardiovasc Med*. (2022) 23(3):185–90. doi: 10.2459/JCM.0000000000001253

9. Orrico M, Ronchey S, Mangialardi N. Coil embolisation of an ascending aortic pseudo aneurysm to avoid a second ascending aortic redo operation. *Eur J Vasc Endovasc Surg*. (2021) 61(2):340. doi: 10.1016/j.ejvs.2020.10.024

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Case report: Unruptured sinus of Valsalva aneurysms causing postural angina pectoris and false "pulmonary hypertension"

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A 37-year-old woman presented with worsening intermittent chest pain and dyspnea in the previous year. Although the dyspnea was exertion-dependent, her chest pain was heavily dependent on her postural position, worsening in the supine position but alleviated by lying prone or by sitting up and leaning forward. She was cyanotic, and a diastolic murmur in the left third intercostal space was auscultated. An electrocardiogram recorded when she laid flat and had angina pectoris attacks showed ST-segment elevation in the aVR and depression in the II, III, aVF, and V3–V6 leads. However, when she sat up for a few minutes, her symptoms and ST-segment abnormalities disappeared. Echocardiography and cardiac computed tomography angiography revealed large unruptured aneurysms of the left and non-coronary sinuses, along with a dilated aortic root, severe aortic regurgitation, and right ventricular high pressure. Coronary angiography showed ~90% pulsating stenosis of the left main coronary artery and ~80% pulsating stenosis of the proximal left circumflex artery, presumably caused by pulsation of the dilated sinus of Valsalva aneurysm under blood pressure. Genetic testing revealed c.1781 C > G nonsense mutations in the *FLNA* gene. The patient underwent surgery, which confirmed dual unruptured left/non-coronary sinus of Valsalva aneurysms. Our case illustrates an unusual postural form of angina pectoris and false "pulmonary hypertension" caused by large dual unruptured left/non-coronary sinus of Valsalva aneurysms.

KEYWORDS

sinus of Valsalva aneurysm, postural angina pectoris, pulmonary hypertension, case report, FLNA gene mutation

Introduction

Sinus of Valsalva aneurysm (SVA) is a rare cardiac abnormality that was first described by Hope in 1839. Its estimated prevalence is 0.09% in the general population (1). It is worth noting that SVA occurs five times more often in Asian than Western populations, with a predominance in men (2). The origin of SVA can be congenital or acquired, with the former being more common. Congenital SVA is caused by defective continuity between the aortic media and aortic valve annulus fibrosis. Previous studies have indicated that mutations in *FBN1* or *MFAP5* may be related to congenital SVA (3). Acquired SVA usually arises from chest trauma, infective endocarditis, syphilis, tuberculosis, aortitis, atherosclerosis, or connective tissue disorders (4). The clinical presentation of SVA varies. Ruptured SVA often causes substernal chest pain, dyspnea, and sudden cardiac arrest, whereas most unruptured SVA cases are asymptomatic (1). Once an unruptured SVA is

identified, surgical repair is vital to prevent its rupture, which can lead to myocardial infarction, malignant arrhythmia, heart failure, and pericardial tamponade. The mean survival period for patients with untreated, ruptured SVA is 3.9 years (5). This report describes a case of dual unruptured left/non-coronary SVAs that led to postural angina pectoris and false "pulmonary hypertension".

Case presentation

A 37-year-old woman presented with worsening intermittent chest pain and dyspnea for 1 year and was admitted to our hospital. Interestingly, although the dyspnea was exertion-dependent, her chest pain heavily depended on the postural position as it worsened in the supine position but was alleviated by lying prone or sitting up and leaning forward. The patient had moderate anemia due to adenomyosis. She had no history of chest trauma, infective endocarditis, syphilis, tuberculosis, hypertension, diabetes, dyslipidemia, aortic aneurysm, or connective tissue disease. Her vital signs were as follows: body temperature 36.5 °C, blood pressure 130/70 mmHg, pulse 75 beats/min, respiratory rate 20 breaths/min, and oxygen saturation 96% (indoor air). A physical examination revealed lip cyanosis (**Figure 1A**) and a diastolic murmur at the left third intercostal space was present. Respiratory and abdominal examinations revealed no abnormal findings. Written informed consent for publication was obtained from the patient.

A hematological evaluation showed a hemoglobin level of 73 g/L (microcytic hypochromic anemia), a white blood cell count of 4.59×10^9/L, and a platelet count of 143×10^9/L. Other laboratory data, including myocardial enzyme levels, NT pro-brain natriuretic peptide levels, coagulation function, and hepatic

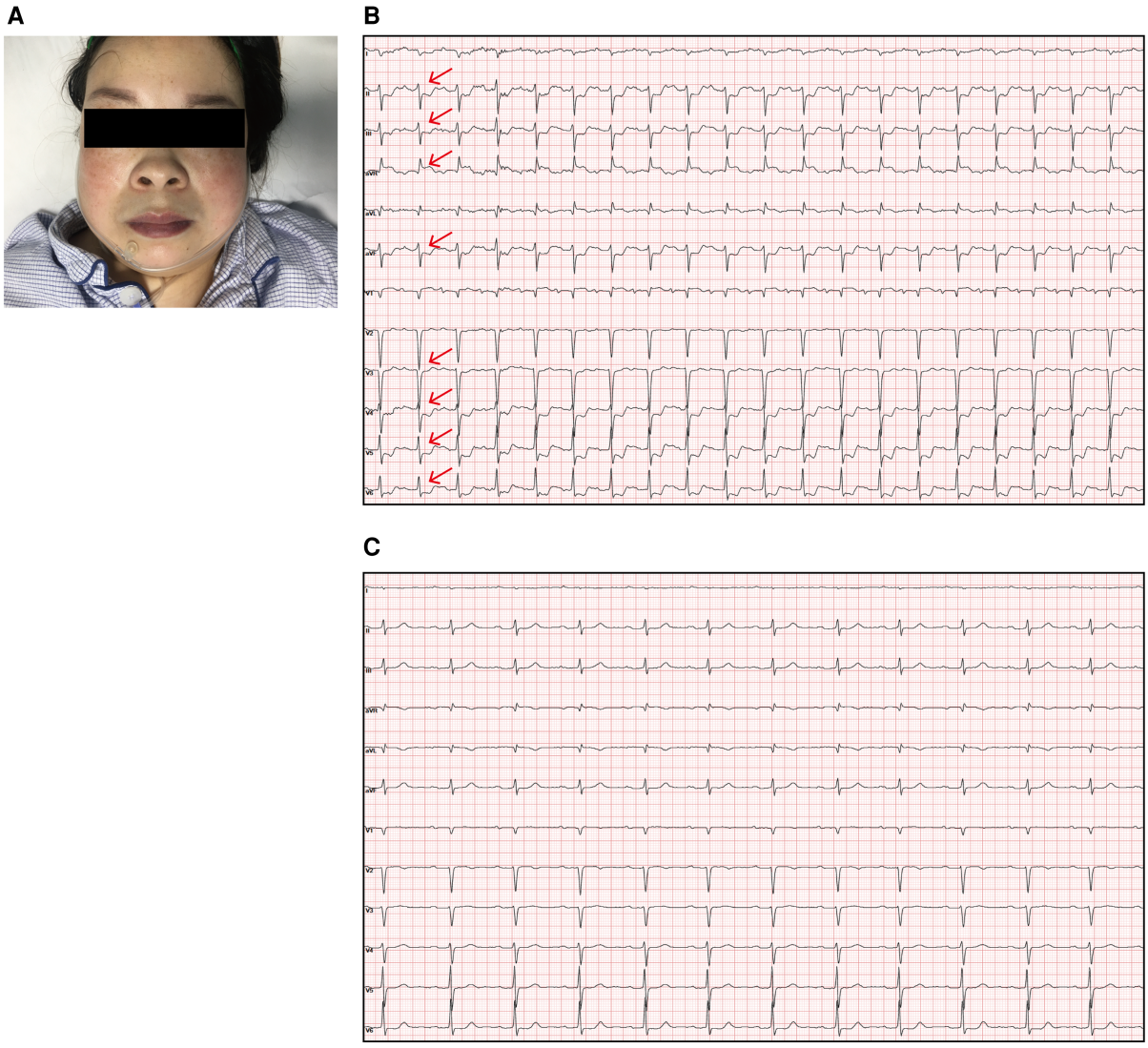

FIGURE 1
(**A**) The patient was cyanotic. (**B**) Electrocardiogram showing sinus rhythm and obvious ST-segment elevation in the aVR (red arrow) as well as depressions in the II, III, aVF, and V3–V6 leads (red arrow) when she laid flat and experienced angina pectoris. (**C**) The angina pectoris and ST segment abnormalities were relieved when she sat up.

and renal function were normal. Electrocardiography showed sinus rhythm, obvious ST-segment elevation in the aVR, and depression in the II, III, aVF, and V3–V6 leads when she lay flat and had angina pectoris attacks (Figure 1B). However, when she sat up for a few minutes, her symptoms and ST segment abnormalities disappeared (Figure 1C). Echocardiography revealed large unruptured aneurysms in both the left (47 mm × 43 mm) and non-coronary (44 mm × 35 mm) sinuses (Figure 2A), along with an enlarged left ventricle (58 mm, Figure 2B), a dilated aortic root (52 mm, Figure 2C), a ventricular septal membranous aneurysm (19 mm × 16 mm, Figure 2D), severe aortic regurgitation (Figure 2E), right ventricular high pressure (pressure gradient 54 mmHg, Figure 2F), and decreased left ventricular systolic function (ejection fraction 51%). Coronary angiography showed ~90% pulsating stenosis of the left main coronary artery (LM) and ~80% pulsating stenosis of the proximal left circumflex artery (LCX), presumably caused by pulsation of the dilated left SVA under blood pressure (Supplementary Video S1). The aortic root was severely distorted when we attempted to intubate the coronary arteries. Non-obstructive atherosclerosis was observed in the right coronary artery (RCA). Cardiac computed tomography angiography (CCTA) clearly showed a large unruptured left SVA (Figure 3A), which was anatomically close to the LM (Figure 3B) and proximal LCX (Figure 3C), causing severe narrowing of the vessel lumens. The dilated left SVA upwardly squeezed the pulmonary trunk (Figure 3D) and accelerated the maximum pulmonary blood flow to 3.7 m/s. The dilated non-coronary SVA adjoined the right atrium without a crevasse (Figure 3A). The right coronary sinus was normal, and the RCA blood flow was smooth. The sagittal plane view showed dual unruptured left/non-coronary SVAs (Figure 3E). Three-dimensional reconstruction images showed the LM and LCX running along the epicardium of the left SVA, with severe narrowing of the LCX (Figure 3F). To rule out other etiologies, C-reactive protein, erythrocyte sedimentation rate, rheumatoid factor, antinuclear antibody, antidouble stranded DNA antibody, anti-neutrophil cytoplasmic antibody, lupus anticoagulant, T-spot, and syphilis serology tests were performed, but all were negative. Genetic testing revealed c.1781 C > G nonsense mutations in the *FLNA* gene. According to the American College of Medical Genetics and Genomics (ACMG) standard, *FLNA* c.1781 C > G was determined as a "likely pathogenic mutation" since it satisfied the criteria of both PVS1 (pathogenic very strong) and PM2 (pathogenic moderate).

On admission, we administered nitroglycerin, an oxygen mask, and metachysis to alleviate her symptoms. Three days later, the patient underwent surgical intervention, including aneurysm patch closure, coronary artery reconstruction, and aortic valve and aortic root replacement. Operative findings confirmed a dual unruptured Valsalva aneurysm of the left coronary and non-coronary sinuses (Figure 4). The right coronary sinus was normal. The modified Bentall procedure was performed using a 26 mm biological composite graft and a mechanical aortic valve (Carbomedics Inc., Austin, TX, United States). The LM artery was then re-implanted into the newly formed left coronary sinus.

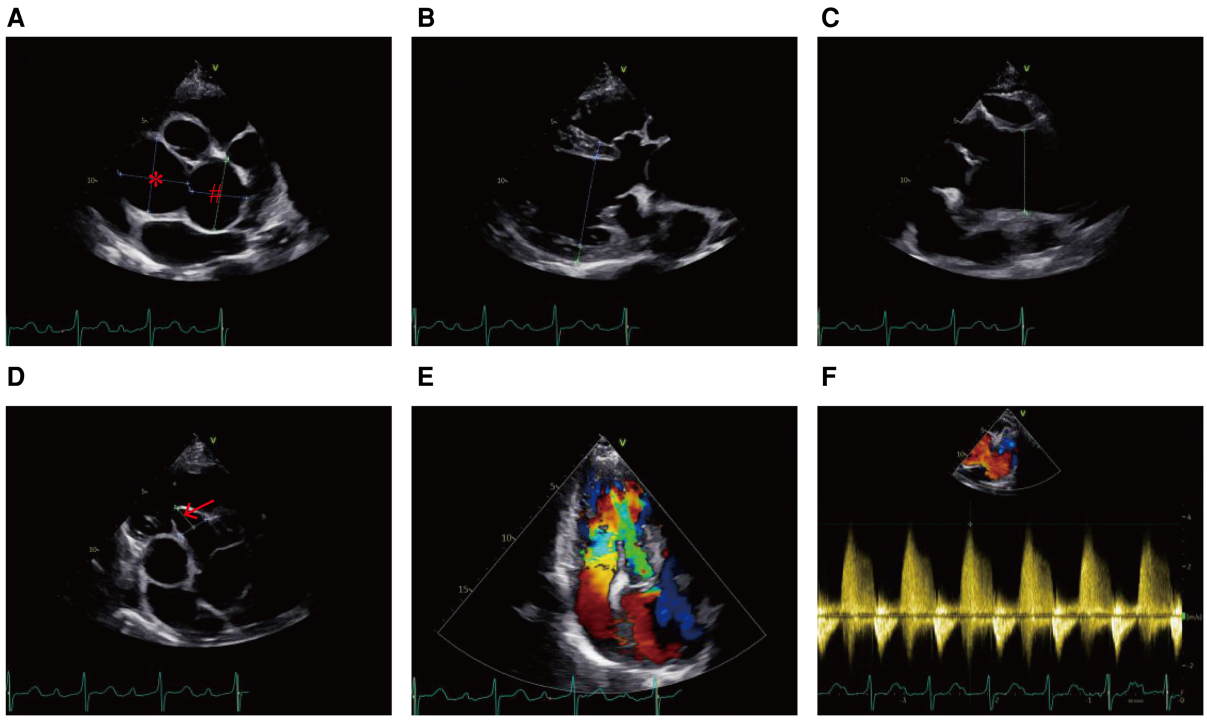

FIGURE 2
Echocardiogram showing (A) the unruptured left SVA (#) and non-coronary SVA (*), (B) enlarged left ventricle, (C) dilated aortic root, (D) ventricular septal membranous aneurysm (red arrow), (E) severe aortic regurgitation, and (F) right ventricular high pressure.

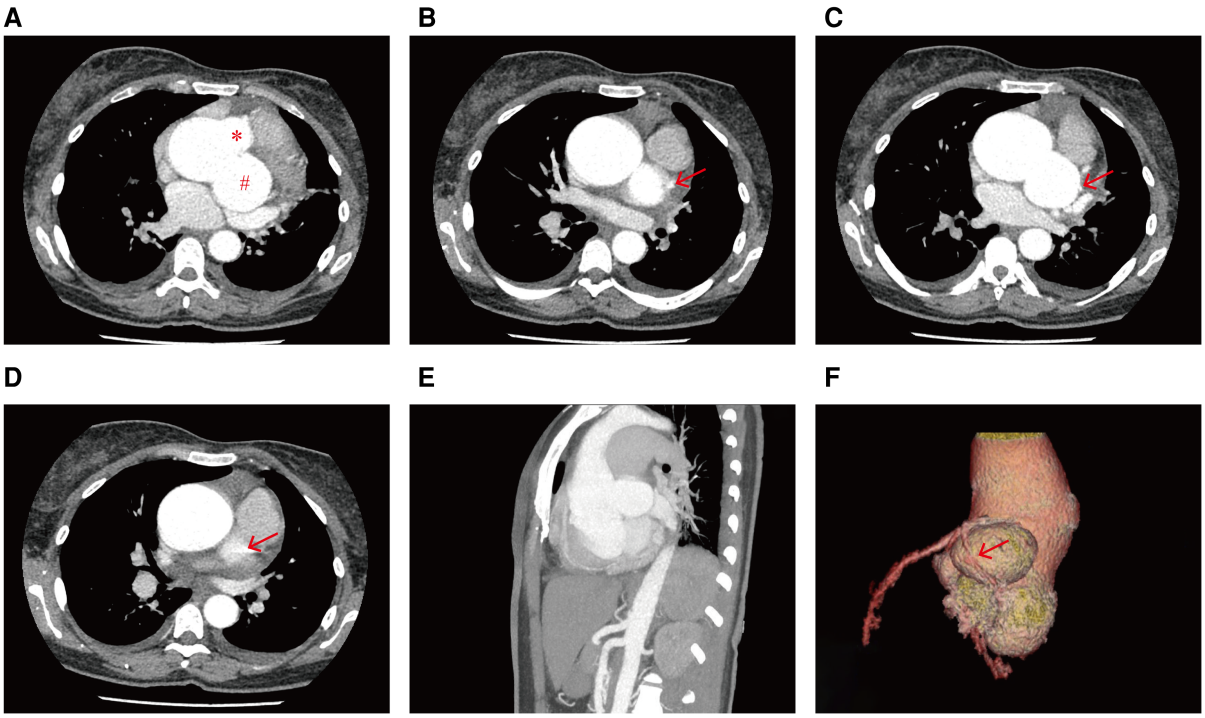

FIGURE 3
CCTA images showing (**A**) the unruptured left SVA (#) and non-coronary SVA (*), the effect of the dilated left SVA on the (**B**) LM (red arrow) and the (**C**) LCX (red arrow), and (**D**) the dilated left SVA upwardly squeezed the pulmonary trunk (red arrow). (**E**) Sagittal plane view of the dual unruptured left/non-coronary SVAs. (**F**) Three-dimensional reconstruction image showing the dual unruptured left/non-coronary SVAs, passage of the LM and LCX, and compression of the LCX (red arrow).

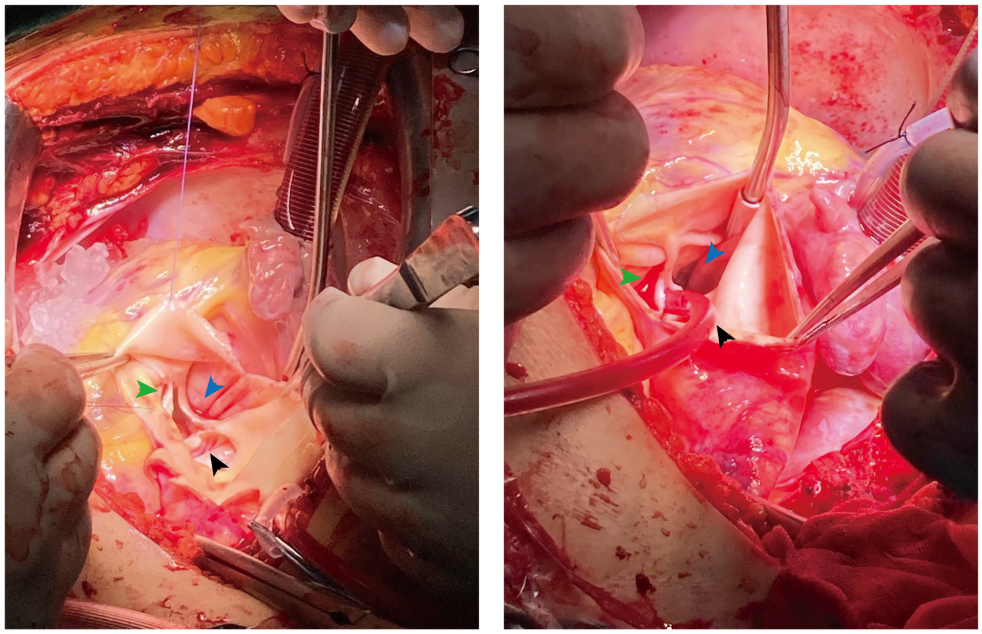

FIGURE 4
Intraoperative images confirmed the dual unruptured left (black arrow)/non-coronary (blue arrow) SVAs. The right coronary sinus (green arrow) was normal.

Unfortunately, the patient experienced cardiac arrest during the operation, and, despite receiving an emergency thoracotomy, the heart was unable to recover after surgery and she died.

Discussion

SVA is an uncommon congenital heart disease and its clinical symptoms depend on its origin and size, its influence on adjacent anatomical structures, and whether it ruptures. Regardless of etiology, the majority of SVA cases originate from the right coronary sinus (65%–86%), followed by the non-coronary sinus (10%–30%), while left coronary sinus aneurysms are rare (2%–5%) (6). However, cases of multiple SVAs are extremely rare (7). In our case of dual unruptured left/non-coronary SVAs, the dilated SVA compressed the LM, LCX, and pulmonary trunk, inducing postural angina pectoris and false "pulmonary hypertension".

The etiology of SVA can be congenital or acquired, with the former being most common. Ventricular septal defects, bicuspid aortic valves, or aortic valve regurgitation often coexist with congenital SVA (8). In this case, echocardiography revealed a dilated aortic root and severe aortic regurgitation, while the aortic valve was tricuspid. Acquired SVA may be accompanied by manifestations of the primary diseases. In our case, none of the acquired pathologies, including chest trauma, infective disease, aortitis, or connective tissue disease, were found. However, a nonsense mutation of the *FLNA* gene, which encodes Filamin A and causes Melnick-Needles Syndrome and Cardiac Valvular Dysplasia, may account for the occurrence of SVA.

In previous studies, unruptured SVAs were generally asymptomatic and often presented as incidental findings during cardiac imaging, whereas those that ruptured into the cardiac chamber or pericardium induced acute heart failure or cardiac tamponade, respectively (9). Right SVAs protrude and rupture into the right ventricle, non-coronary SVAs tend to rupture into the right atrium, and left SVAs typically rupture into the pulmonary artery, left ventricle, or pericardium (10). Hemodynamic alterations can lead to relevant manifestations. Blood stagnation in the SVA leads to embolization, which may migrate the coronary or peripheral arteries (11). Several reports have described that left or right SVAs cause compression of the LAD, while non-coronary SVAs cause compression of the RCA, which results in angina pectoris (9, 12, 13). In our patient, the dilated left SVA compressed the LM and proximal LCX during systole, inducing acute interruption of coronary blood flow, which was relieved during diastole. Her symptoms were alleviated when she sat up due to the change in the direction of the effect of gravity on the left SVA. Dynamic electrocardiographic changes suggested an intrinsic mechanism: a mass effect of the left SVA on the LM and LCX since they run along the epicardium of the left SVA. In the supine position, the dilated left SVA compressed the epicardial coronary artery, impairing blood flow. However, the mass effect of the left SVA was relieved in the prone or upright and seated positions. CCTA clearly showed that the LM and proximal LCX narrowed and ran along the dilated left SVA. In the majority of relevant studies, coronary ischemia was caused by SVA compression; however, this study is the first to report that these symptoms are posture-dependent.

Patients with congenital heart diseases who develop pulmonary hypertension are always affected by left-to-right shunting such as atrial or ventricular septal defects. A ruptured right SVA fistulizing into the right atrium caused reversible flow-induced pulmonary hypertension (14). Mansour et al. (15) reported a very large right SVA compressing the right ventricular outflow tract that caused high right intraventricular pressure and tricuspid regurgitation in an elderly man. In our case, the dilated left and non-coronary SVAs did not rupture into the right heart system, and no crevasse was noted in the ventricular septum. Unusually, the unruptured left SVA grew sufficiently large to squeeze the pulmonary trunk, thus decreasing the pulmonary blood flow volume and pulmonary oxygenation. This is a case of false "pulmonary hypertension" in this patient as the right ventricular high pressure was caused by extrinsic compression of the pulmonary trunk. The oxygen-rich blood flow from the pulmonary capillaries to the peripheral arteries through the left heart was insufficient, which caused dyspnea and cyanosis.

The diagnosis and assessment of SVA relies on echocardiography, CCTA, and MRI (9). Three-dimensional images allow for a more accurate evaluation of aneurysms within the cardiac chambers and coronary arteries. Invasive angiography is risky in patients with SVA as their coronary arteries are difficult to engage, and their aortic roots are more prone to injury. Based on surgical series, case reports, and single-center data, early or emergent surgical repair is recommended in symptomatic unruptured and ruptured patients (16). As the unruptured SVAs caused postural angina pectoris and dyspnea in this case, we decided on surgical intervention immediately after systematic examination. Patch closure reduces the SVA volume, and aortic valve replacement or valvuloplasty is necessary in cases of aortic valve regurgitation. Other incisions are performed when the SVA ruptures into the cardiac chambers. Coronary artery bypass grafting should be performed in cases of an SVA-compressed epicardial coronary artery. Reparative surgery is associated with a low perioperative mortality rate, a low risk of recurrence, and long-term survival (5). Percutaneous aneurysm closure devices have been shown to be an alternative treatment with promising results (17).

Unfortunately, our patient experienced cardiac arrest during the operation. Several issues might have caused the death: (1) the dilated left SVA might have severely compressed the coronary arteries during anesthesia and thoracotomy, which might have induced acute myocardial infarction. Inadequate cardiac output might have led to circulatory collapse. (2) The pulmonary trunk was completely obstructed by the SVA. There was no oxygen-rich blood coming from the pulmonary circulation to the heart, which might have induced cardiac arrest and oxygen depletion. (3) Anatomically, the atrioventricular node is close to the non- and right-coronary sinuses. Electrocardiography revealed 1st atrioventricular block in this patient on admission. We presume that the non-coronary SVA might have compressed the atrioventricular node and might have induced complete atrioventricular block during the operation.

In conclusion, SVA is a rare cardiac abnormality with several clinical manifestations. This case suggests that, in patients with unexplained postural angina pectoris or pulmonary hypertension, the possibility of SVA should be considered. Echocardiography should be immediately performed in suspected cases. A prompt diagnosis can be established based on CCTA findings. Once identified, immediate surgical intervention as soon as possible is recommended.

Data availability statement

The original contributions presented in the study are included in the article/**Supplementary material**, further inquiries can be directed to the corresponding author.

Ethics statement

Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

Author contributions

CZ collected the study sample and wrote the manuscript. JT provided clinical information. SL designed and organized the study. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

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References

1. Weinreich M, Yu PJ, Trost B. Sinus of Valsalva aneurysms: review of the literature and an update on management. *Clin Cardiol.* (2015) 38(3):185–9. doi: 10.1002/clc.22359

2. Chu SH, Hung CR, How SS, Chang H, Wang SS, Tsai CH, et al. Ruptured aneurysms of the sinus of Valsalva in oriental patients. *J Thorac Cardiovasc Surg.* (1990) 99(2):288–98. doi: 10.1016/S0022-5223(19)37013-8

3. Milewicz DM, Regalado E. Heritable thoracic aortic disease overview. In: MP Adam, HH Ardinger, RA Pagon, SE Wallace, LJH Bean, G Mirzaa, et al. editors. Genereviews® [Internet]. Seattle WA: University of Washington, Seattle (1993).

4. Lindsay AC, Mahesh B, Gaer JA, Dalby MC. Ruptured sinus of Valsalva aneurysm presenting as ST-elevation myocardial infarction. *Nat Rev Cardiol.* (2009) 6(5):379–82. doi: 10.1038/nrcardio.2009.45

5. Sarikaya S, Adademir T, Elibol A, Buyukbayrak F, Onk A, Kirali K. Surgery for ruptured sinus of Valsalva aneurysm: 25-year experience with 55 patients. *Eur J Cardiothorac Surg.* (2013) 43(3):591–6. doi: 10.1093/ejcts/ezs450

6. Galicia-Tornell MM, Marin-Solis B, Mercado-Astorga O, Espinoza-Anguiano S, Martinez-Martinez M, Villalpando-Mendoza E. Sinus of Valsalva aneurysm with rupture. Case report and literature review. *Cir Cir.* (2009) 77(6):441–5.

7. Chigurupati K, Kumaresan B, Gadhinglajkar S, Sreedhar R. Multiple unruptured aortic sinus of Valsalva aneurysms: a rare presentation. *Echocardiography.* (2017) 34 (2):317–9. doi: 10.1111/echo.13423

8. Daud E, Or T, Adler Z, Shturman A. The value of three-dimensional echocardiography in the diagnosis and management of ruptured sinus of Valsalva aneurysm: a case report. *Eur Heart J Case Rep.* (2019) 3(3):ytz111. doi: 10.1093/ehjcr/ytz111

9. Dias RR, Camurca FD, Leite Filho OA, Stolf NA. Right sinus of Valsalva aneurysm causing extrinsic coronary compression. *Arq Bras Cardiol.* (2009) 92(6):e47–50. doi: 10.1590/s0066-782x2009000600017

10. Simonavicius J, Jarasuniene D. Angina pectoris and sinus of Valsalva aneurysm: a rare manifestation of an uncommon disease. *Acta Med Litu.* (2018) 25(4):234–9. doi: 10.6001/actamedica.v25i4.3934

11. Ponti A, Qanadli SD, Kirsch M, Tapponnier M. Left sinus of Valsalva aneurysm as a cause of chronic stable angina. *Interact Cardiovasc Thorac Surg.* (2017) 24 (6):967–8. doi: 10.1093/icvts/ivx041

12. Saraswat A, Malawaraarachchi N, Stroebel A, Jayasinghe R. Sinus of Valsalva aneurysm causing compression of the left anterior descending artery resulting in angina. *JACC Cardiovasc Interv.* (2019) 12(17):e141–3. doi: 10.1016/j.jcin.2019.05.030

13. Iwakawa H, Iino T, Yamamoto H, Watanabe H. Large non-coronary sinus of Valsalva aneurysm: a case report of an unusual cause of angina pectoris. *Eur Heart J Case Rep.* (2020) 4(5):1–6. doi: 10.1093/ehjcr/ytaa241

14. Al-Sabeq B, De S, Davey R. A case of ruptured sinus of Valsalva aneurysm and reversible flow-induced pulmonary hypertension. *Pulm Circ.* (2018) 8 (2):2045894018760656. doi: 10.1177/2045894018760656

15. Mansour MJ, Mansourati J, Le Ven F, Didier R, Gilard M, Jobic Y. Giant sinus of Valsalva aneurysm mimicking severe pulmonary hypertension. *Int J Cardiovasc Imaging.* (2020) 36(9):1695–6. doi: 10.1007/s10554-020-01875-9

16. Hiratzka LF, Bakris GL, Beckman JA, Bersin RM, Carr VF, Casey DE Jr, et al. 2010 ACCF/AHA/AATS/ACR/ASA/SCA/SCAI/SIR/STS/SVM guidelines for the diagnosis and management of patients with thoracic aortic disease: a report of the American college of cardiology foundation/American heart association task force on practice guidelines, American association for thoracic surgery, American college of radiology, American stroke association, society of cardiovascular anesthesiologists, society for cardiovascular angiography and interventions, society of interventional radiology, society of thoracic surgeons, and society for vascular medicine. *Circulation.* (2010) 121(13):e266–369. doi: 10.1161/CIR.0b013e3181d4739e

17. Liu S, Xu X, Zhao X, Chen F, Bai Y, Li W, et al. Percutaneous closure of ruptured sinus of Valsalva aneurysm: results from a multicentre experience. *EuroIntervention.* (2014) 10(4):505–12. doi: 10.4244/EIJV10I4A87

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Noblestitch® system for PFO closure: A novel but judicious alternative to traditional devices—A case report

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Background: Percutaneous suture-mediated patent foramen ovale (PFO) closure has recently been used with the aim of avoiding double-disc nitinol device implantation. This novel technique has been carried out successfully in several centers offering PFO closure with an effective closure rate comparable to conventional double-disc devices.

Case summary: A 50-year-old man, a pentathlon athlete, suffering from a previous left-sided ischemic stroke, underwent percutaneous closure of a permanent right-to-left shunt *via* PFO with a large fenestrated septum primum aneurysm at another institution. The NobleStitch® system was successfully implanted using local anesthesia and under angiographic-fluoroscopic monitoring. He was discharged home on aspirin 100 mg daily with a moderate residual shunt on contrast transthoracic echocardiography (cTTE) that persisted unaltered at subsequent controls. After 7 months, unable to resume sporting activity because of physical discomfort and dyspnea on exertion, the patient asked for a second opinion at our Heart and Brain clinic. Two-dimensional (2D) TTE showed septum primum laceration next to a radiopaque polypropylene knot with a moderate bidirectional shunt located at the fenestrated septum primum far from the PFO site. A catheter-based closure of the septal defect was therefore planned under local anesthesia and rotational intracardiac echo monitoring. An equally sized discs 28.5 mm × 28.5 mm Flex II UNI occluder (Occlutech GmbH, Jena, Germany) was successfully implanted across the atrial septal defect without complications. The patient was discharged in good clinical conditions; dual antiplatelet therapy (aspirin 100 mg/daily and clopidogrel 75 mg/daily) was recommended for 2 months and then single antiplatelet therapy (aspirin100 mg/daily) up to 6 months. Abolition of the residual shunt was confirmed at 1- and 6-month follow-up by contrast transcranial Doppler and 2D color Doppler cTTE.

Discussion: Closing a PFO with a suture-base system, without leaving a device implant behind, may be a cutting-edge technology and potential alternative to traditional devices. Nevertheless, meticulous selection of the PFO anatomies by 2D TEE is key to a successful closure procedure in order to avoid complications that must be managed again with a second percutaneous procedure or by surgery.

KEYWORDS

patent foramen ovale, transcatheter closure, suture-based PFO closure, atrial septal tear, residual shunt

Introduction

In carefully selected patients with cryptogenic stroke, patent foramen ovale (PFO) closure has been shown to significantly reduce the risk of recurrent neurological events and new brain infarcts compared with antiplatelet treatment alone (1–3).

Conventional PFO nitinol double-disc occluders, routinely used for this purpose, are associated with potential but extremely low risk of early and late complications (arrhythmias, thrombus formation, embolization, erosion). Among the limitations of traditional occluders, future transseptal puncture and left-sided interventions may be impeded by the presence of a bulky device covering the interatrial septum.

Recently, a percutaneous suture-mediated system to close the PFO has been added to interventional practice with encouraging results in terms of safety and efficacy, as long as a meticulous selection of suitable PFO anatomies is carried out. Currently, closure rates with the suture-mediated system appear to be similar to those obtained with double-disc metal-alloy occluders (4). Nevertheless, there is still a paucity of data describing in detail the efficacy and safety of the NobleStitch® system.

Case presentation

A 50-year-old man, a professional pentathlon athlete without documented risk factors for heart disease, suffered from a left-sided ischemic stroke with dysarthria and right hemianopsia. He was admitted to the stroke unit of another institution where brain magnetic resonance imaging (MRI) showed left posterior insular cortex ischemic stroke. Thrombolysis with intravenous tissue plasminogen activator (IV-tPA) was administered. Afterward, contrast transthoracic and transesophageal echocardiography (cTTE/TEE) and contrast Transcranial Doppler (cTCD) were performed at the Cardiology Department of this institution confirming "spontaneous severe right-to-left shunt (RLS) via a PFO 17.5 mm in length, >5 mm in width associated with abnormally redundant fenestrated septum primum aneurysm (ASA) protruding predominantly to the right atrium in basal conditions and to the left atrium during the cardiorespiratory cycle" (**Supplementary Figure S1**). Notwithstanding, the PFO anatomy was considered appropriate by the local team, and it was decided to implant a suture-based "deviceless" NobleStitch® EL system, consisting of two polypropylene sutures (one for the septum primum and one for the septum secundum) tightened together by a dedicated delivery and sealing system (KwiKnot). The patient was discharged home on aspirin 100 mg daily for 1 month. Two-dimensional (2D) TTE color Doppler at discharge showed a moderate residual bidirectional shunt, primarily left-to-right across the septum primum fenestration (septal defect), that persisted unaltered on subsequent controls. After 7 months, the patient was unable to resume his sporting activity and attended our Heart and Brain clinic for a second opinion due to tiredness, dyspnea on exertion, and persistent mild dysarthria, a sequela of the index event. Luckily, no new lesions were found at the control brain MRI. Nevertheless, 2D TTE color Doppler confirmed the redundant ASA, the small residual shunt across the tear next to the radiopaque polypropylene knot, and the persistent predominant left-to-right shunt through the atrial septal defect located far from the PFO site. After the heart team discussion, written informed consent was obtained from the patient. A catheter-based atrial septal defect closure was planned under local anesthesia and rotational intracardiac echo (Ultra ICE, Boston Scientific Corporation, San Jose, CA, United States) monitoring, the standard procedural guidance at our center. The tearing of the aneurysmatic septum primum, next to the KwiKnot and probably induced by the stretching of the NobleStitch® device, and the septal defect were clearly visible by rotational intracardiac echo (**Figures 1A–C**). In order to anchor the device on septum secundum rims, a Flex II UNI occluder (Occlutech GmbH, Jena, Germany) with equally sized discs (28.5 mm × 28.5 mm) was successfully implanted under intracardiac echo (**Figures 2A–C**) and fluoroscopic guidance (**Figures 3A–F**) across the septal defect, "sandwiching" the KwiKnot between the two discs and covering the septum primum laceration next to the knot. Intraprocedural agitated saline injection from the femoral vein confirmed abolition of the interatrial residual shunt (**Figure 2D**). The patient was discharged home the following day in good clinical conditions. Dual antiplatelet therapy (aspirin 100 mg/daily and clopidogrel 75 mg/daily) was recommended for the first 2 months and then single antiplatelet therapy (aspirin 100 mg/daily) up to 6 months.

cTTE and cTCD at 1- and 6-month follow-up confirmed the stable position of the nitinol double-disc device without residual shunt (**Figure 4**). Currently, the dysarthria has improved, and the patient has resumed competitive sporting activity 2 months after the procedure.

Discussion

The concept of closing a PFO with a suture-based system, without leaving a device implant behind, is very innovative and a potential alternative to nitinol double-disc devices. The NobleStitch® system closes the PFO by applying sutures through the septum primum and septum secundum, subsequently creating a knot between the two sutures, and removing excess suture material. Only one registry has been published (4) with effective PFO closure in the majority of septal anatomies without complications. Thereafter, a retrospective observational study of 247 patients sought to assess PFO anatomy by preprocedural TEE in patients deemed suitable for suture-mediated technique to identify predictors of the most common complication, a post-procedural residual shunt (5). This study revealed that PFO > 5 mm in width and spontaneously large RLS are less likely to be closed with one stitch only. More recently, another prospective single-center study with a 6-month follow-up period ($n = 116$) investigated factors that may have contributed to residual intracardiac shunting ≥2 (20%, $n = 23$), revealing that the

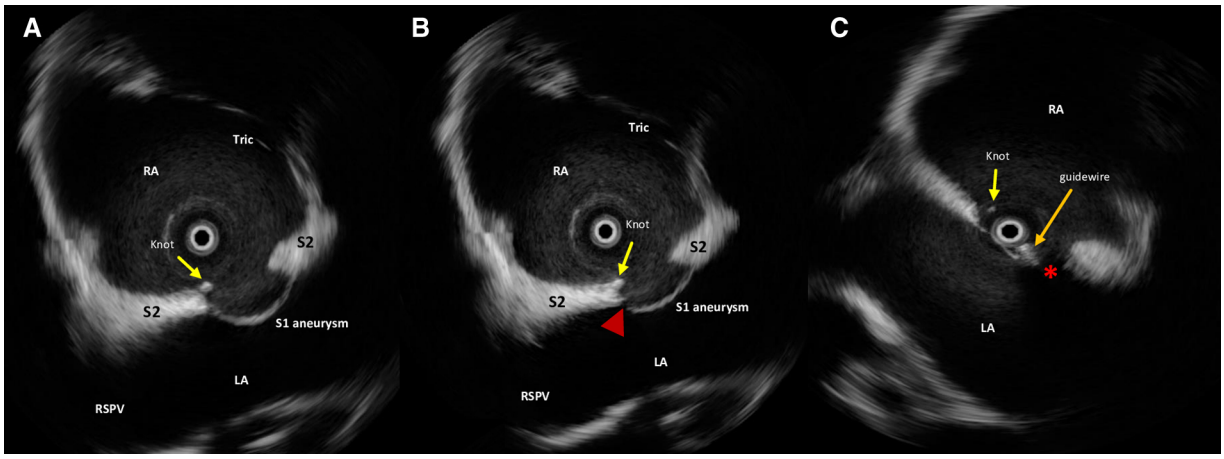

FIGURE 1
Intraprocedural rotational intracardiac Echo by Ultra ICE (mechanical 9F/9 MHz 360° scan probe, Ultra ICE™, EP technologies, BSC, CA, United States) in the parasagittal long axis ("four chamber") plane showing septum primum aneurysm, NobleStitch EL sealing system (radiopaque polypropylene knot, yellow arrow), laceration of the septum primum next to the knot (red arrowhead) (**A**,**B**) and the guidewire (orange arrow) crossing the septal fenestration (red asterisk) (**C**). S1, septum primum; S2, septum secundum; RA, right atrium; LA, left atrium; RSVP, right superior pulmonary vein; Tr, tricuspid valve.

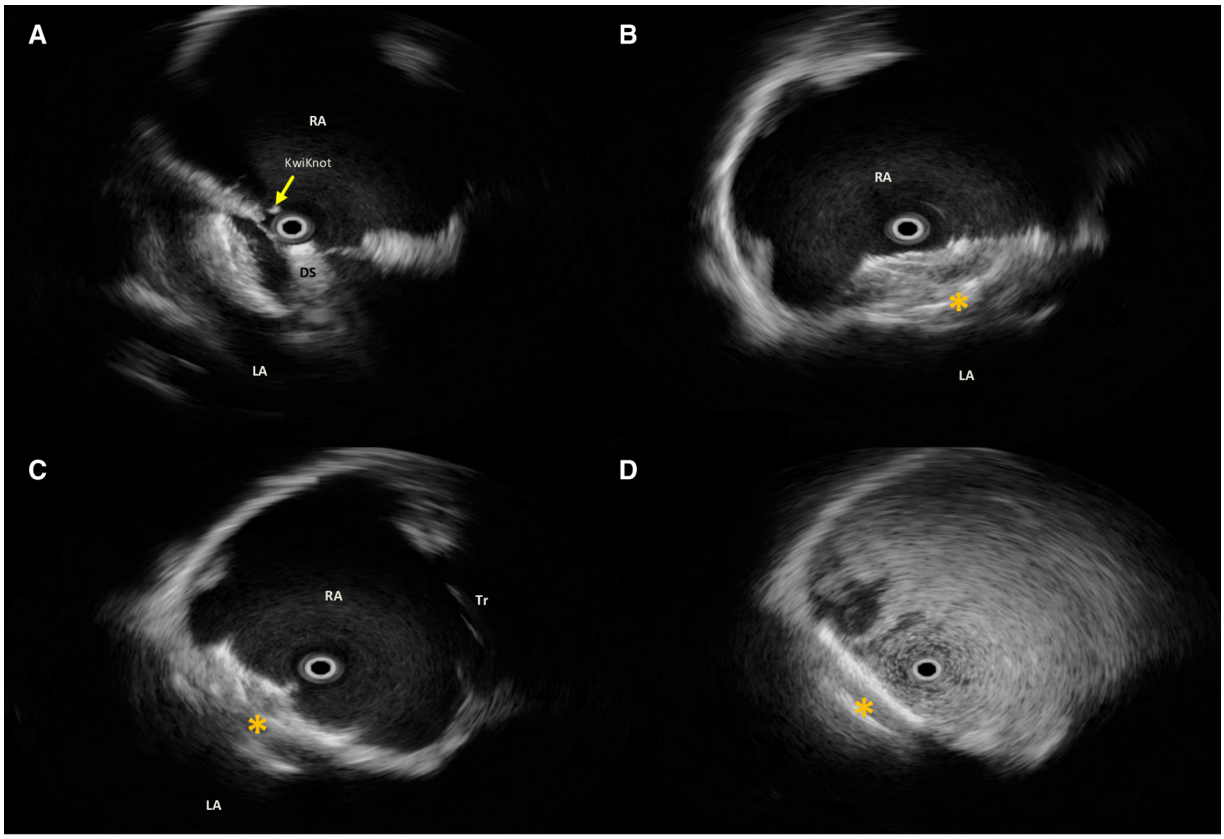

FIGURE 2
Intraprocedural rotational intracardiac Echo by Ultra ICE (mechanical 9F/9 MHz 360° scan probe, Ultra ICE™, EP technologies, BSC, CA, United States) in the parasagittal long axis ("four chamber") plane showing procedural steps (**A**–**C**) of implantation of Flex II UNI occluder (Occlutech GmbH, Jena, Germany) with equally sized discs (28.5 mm × 28.5 mm) (orange asterisk); (**D**) after agitated saline solution injection from the femoral vein, no residual shunt immediately after device release. RA, right atrium; LA, left atrium; DS, delivery sheath across the septal fenestration.

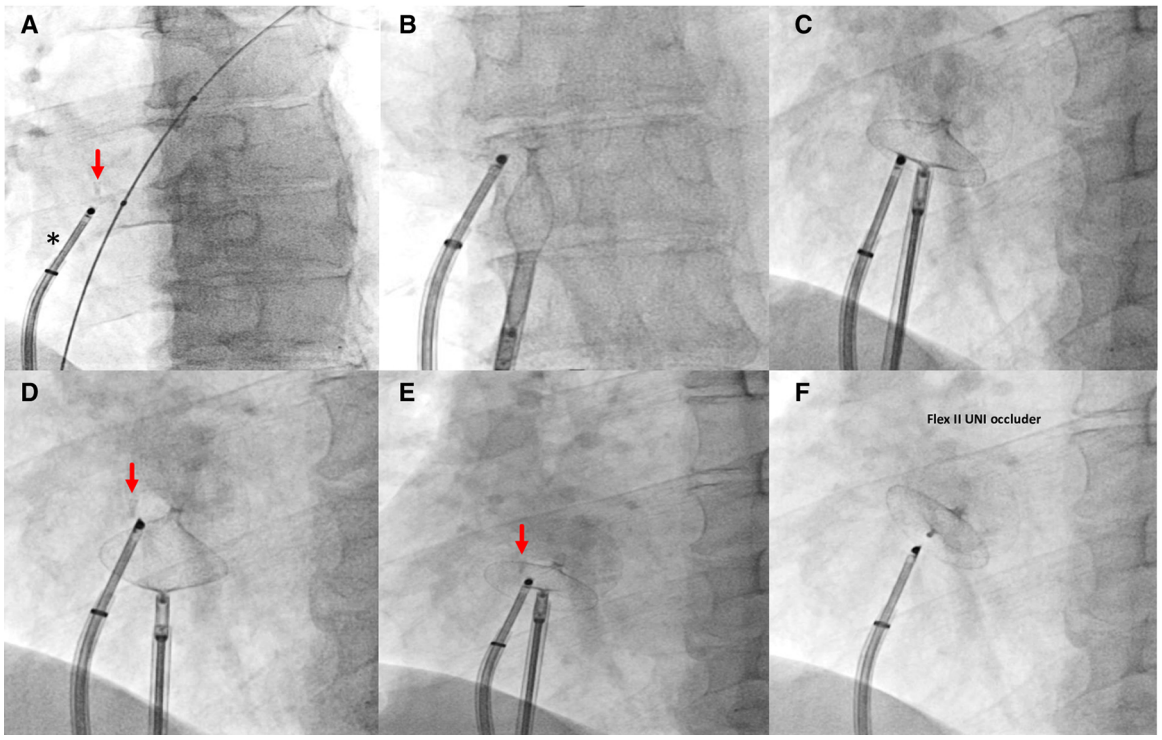

FIGURE 3
Fluoroscopic steps of implantation of Flex II UNI occluder (Occlutech GmbH, Jena, Germany) with equally sized discs (28.5 mm × 28.5 mm) under rotational intracardiac echo by Ultra ICE (black asterisk). (**A–C**) left and right disc opening; (**D,E**) pull and push maneuver to test device stability; (**F**) device deployed. Red arrow, radiopaque polypropylene knot.

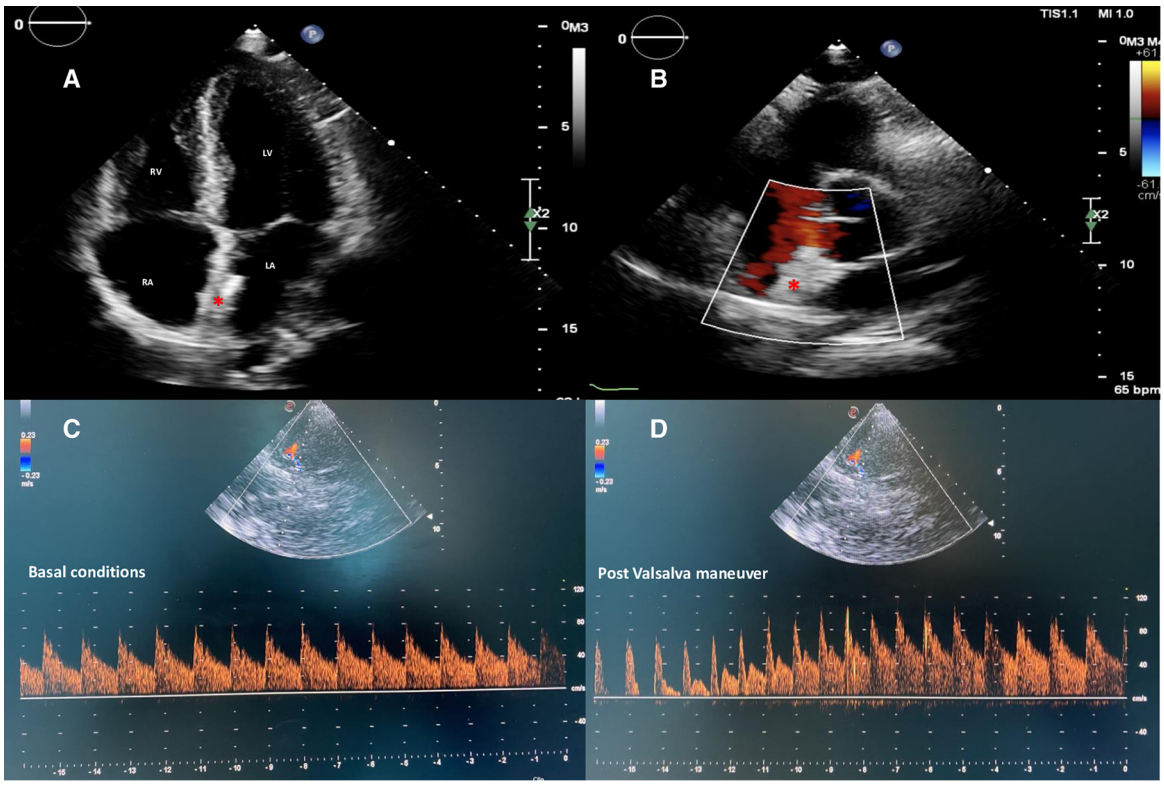

FIGURE 4
Two-dimensional TTE color Doppler in apical four-chambers (**A**) and parasternal short-axis (**B**) views showing a correct and stable position of the equally sized discs nitinol device (red asterisk) without residual shunt. cTCD confirming abolition of the interatrial shunt both in basal conditions (**C**) and after adequate Valsalva maneuver (**D**). TTE, transthoracic echocardiography; cTCD, contrast transcranial Doppler.

principal causes were partial stitch detachment ($n = 12$), atrial septal tear ($n = 3$), and KwiKnot embolization ($n = 2$) (6).

Moreover, a modified technique involving double suture of the septum primum resulted in improved efficacy of the procedure by increasing the surface contact between the septa without the need to place a second septum secundum stitch. However, the current findings are limited by the few cases included in the report (7).

Finally, the suture-based "deviceless" NobleStitch® system has not been gradually growing in importance, and there is limited experience worldwide. On the other hand, an increasing number of reports have documented iatrogenic septum primum lacerations resulting from the use of the NobleStitch® system. These lacerations can cause persistent residual shunts that do not reduce or disappear over time and, as a result, require management, either with a second percutaneous procedure using conventional devices or by surgery (8–10).

Lastly, a PFO Comparative Trial (STITCH) is recruiting patients in the US and European Union with the aim of comparing the effective closure rate of NobleStitch EL vs. FDA Approved Amplatzer Occluder Device (ClinicalTrials.gov identifier: NCT04339699).

It is noteworthy to mention that longer follow-up is imperatively justified to clarify not only the efficacy, but also the safety of this novel but selective alternative to traditional devices.

Conclusion

If PFO closure to prevent recurrent cryptogenic stroke is here to stay, it is imperative to avoid any residual shunts during mid- and long-term follow-up. While suture-based PFO closure systems offer a promising alternative to conventional occluders, a meticulous patient selection process is essential. Specifically, it is necessary to exclude unsuitable PFO anatomies, such as a prominent and fenestrated ASA, PFO > 5 mm in width, septum secundum lipomatous hypertrophy, and spontaneously large RLS, to achieve procedural success and reduce the risk of new embolic events.

Data availability statement

The original contributions presented in the study are included in the article/**Supplementary Material**, further inquiries can be directed to the corresponding author.

Ethics statement

The studies involving human participants were reviewed and approved by Scientific Institute for Research, Hospitalization and Healthcare (I.R.C.C.S.) Ospedale Galeazzi-Sant'Ambrogio, GSD, Milan, Italy Milan, Italy. The patient provided his written informed consent to participate in this study. Written informed consent was obtained from the patient for the publication of any potentially identifiable images or data included in this article.

Author contributions

Conceptualization: EO and AB; methodology: LG and EO; software: GM and AM; validation: EO; formal analysis: AB and EO; investigation: FF, GM, and EO; writing—original draft preparation: EO; writing—review and editing: AB, AM, and EO; visualization: EO; supervision: EO and AB; data curation: GM, AM, and LG. All authors contributed to the article and approved the submitted version.

Conflict of interest

EO is consultant for Occlutech, manufacturer of the device.

The remaining authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fcvm.2023.1095661/full#supplementary-material.

References

1. Mas JL, Derumeaux G, Guillon B, Massardier E, Hosseini H, Mechtouff L, et al. Patent foramen ovale closure or anticoagulation vs. antiplatelets after stroke. *N Engl J Med*. (2017) 377(11):1011–21. doi: 10.1056/NEJMoa1705915.

2. Saver JL, Carroll JD, Thaler DE, Smalling RW, MacDonald LA, Marks DS, et al. Long-term outcomes of patent foramen ovale closure or medical therapy after stroke. *N Engl J Med*. (2017) 377(11):1022–32. doi: 10.1056/NEJMoa1610057.

3. Søndergaard L, Kasner SE, Rhodes JF, Andersen G, Iversen HK, Nielsen-Kudsk JE, et al. Patent foramen ovale closure or antiplatelet therapy for cryptogenic stroke. *N Engl J Med*. (2017) 377(11):1033–42. doi: 10.1056/NEJMoa1707404

4. Gaspardone A, De Marco F, Sgueglia GA, De Santis A, Iamele M, D'Ascoli E, et al. Novel percutaneous suture-mediated patent foramen ovale closure technique: early results of the NobleStitch EL Italian registry. *EuroIntervention*. (2018) 14: e272–9. doi: 10.4244/EIJ-D-18-00023.

5. Gaspardone A, Sgueglia GA, De Santis A, D'Ascoli E, Iamele M, Piccioni E, et al. Predictors of residual right-to-left shunt after percutaneous suture-mediated patent fossa ovalis closure. *JACC Cardiovasc Interv*. (2020) 13:2112–20. doi: 10.1016/j.jcin.2020.06.004.

6. Zannoni J, Popolo Rubbio A, Tusa MB, Corciu AI, Casenghi M, Cannone G, et al. Mechanisms of ineffective patent foramen ovale closure using the percutaneous suture-mediated NobleStitch system. *EuroIntervention*. (2022) 18(1):68–70. doi: 10.4244/EIJ-D-21-01021.

7. Gaspardone A, De Santis A, Giannico MB, Sgueglia GA. Modified percutaneous suture-mediated patent fossa ovalis. *Catheter Cardiovasc Interv*. (2020) 96:638–42. doi: 10.1002/ccd.28936.

8. Baldetti L, Ferri LA, Ancona M, Bellini B, Visco E, Melillo F, et al. Interatrial septal tear after patent foramen ovale closure with the NobleStitch device. *JACC: Cardiovasc Interv*. (2019) 12(16):e139–140. doi: 10.1016/j.jcin.2019.05.040.

9. Trabattoni D, Gili S, Teruzzi G, Tamborini G. A severe right-to-left intracardiac shunt after NobleStitch failure: when a device is needed. *Eur Heart J Case Rep*. (2020) 4:1–4. doi: 10.1093/ehjcr/ytaa162.

10. Gaetti G, Beneduce A, La Fauci D, Scardoni A, Chiappa F, Bellini L, et al. Suture-mediated patent foramen ovale closure using the NobleStitch EL: results from a hospital-based HTA. *Int J Environ Res Public Health*. (2022) 19:7863. doi: 10.3390/ijerph19137863.

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Case report: Stenosis turned leak … and turned stenosis—complications of paravalvular prosthetic leak closure with a plug device

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Background: Paravalvular leak is one of the most common complications and is among the most important prognostic factors of short- and long-term mortality after transcatheter aortic valve implantation (TAVI). Percutaneous valvular leak repair constitutes a first-line treatment for paravalvular leaks and is associated with high success rates and few serious complications nowadays. To the best of our knowledge, this is the first case where placement of the device through the stenting of the bioprosthesis resulted in creating a new symptomatic stenosis that required surgery.

Case summary: We present a case of a patient with low-flow, low-gradient aortic stenosis treated with transfemoral implantation of a biological aortic prosthesis. One month after the procedure, the patient presented with acute pulmonary oedema and a paravalvular leak was discovered, which was corrected by percutaneous repair with a plug device. Five weeks after the valvular leak repair, the patient was readmitted for heart failure. At this time, a new aortic stenosis and paravalvular leak were diagnosed and the patient was referred for surgery. The new aortic mixed diseased was caused by the positioning of the plug device through the valve's metal stenting, which resulted in a paravalvular leak and pressed against the valve's leaflets, causing valvular stenosis. The patient was referred for surgical replacement and evolved well afterward.

Conclusion: This case illustrates a rare complication of a complex procedure, and it highlights the need for multidisciplinary decisions and good cooperation between the cardiology and cardiac surgery teams to develop better criteria in the selection of the appropriate technique for managing paravalvular leaks after TAVI.

KEYWORDS

percutaneous valve therapy, paravalvular leak repair, aortic valve disease percutaneous intervention, transcatheter aortic valve implantation (TAVI), case report

Introduction

Despite the broadening of indications for transcatheter aortic valve implantation (TAVI) to include low-risk patients, as supported by subsequent studies (1, 2), some complications still undermine the use of this technique. Paravalvular leak (PVL) is one of the most common complications and is amongst the most important prognostic factors of

mortality at short- and long-term after TAVI (3–5), being associated with a threefold increase in 30-day mortality (95% CI: 1.73–5.02) and a 2.3-fold increase in 1-year mortality (95% CI: −1.84 to 2.81) for moderate to severe leaks (6).

When comparing surgical to percutaneous aortic valve replacement, the incidence of moderate to severe paravalvular leaks between the percutaneous and the surgical series did not differ significantly in the PARTNER trial: the percutaneous group presenting 0.6% and the surgical group 0.5% at 1 year. By contrast, mild paravalvular leak at 1 year is still significantly higher in the percutaneous series, with 29.4% compared to 2.1% in the surgical one (2).

Here, we present the first case of paravalvular leak after TAVI treated percutaneously with an *Amplatzer* device where migration of the device resulted in severe aortic stenosis needing a surgical intervention.

Case presentation

A 79-year-old female with a history of hypertension, permanent atrial fibrillation, and progressing aortic stenosis presented with NYHA stage II dyspnoea and peripheral oedema. Echocardiography showed a tricuspid aortic valve with severe paradoxical low-flow, low-gradient aortic stenosis with a surface of 0.5 cm^2 by planimetry, a mean gradient of 19 mmHg, a preserved ejection fraction at 60%, and a low left ventricular output of 25 $ml/min/m^2$ due to moderate hypertrophy. A CT scan revealed a modified Agatston calcium score of 630, with a calcium volume of 197 mm^3 and moderate calcifications of the valve with heterogenous peripheral distribution. The patient's surgical risk was characterised by an EuroSCORE II of 1.60% and an STS score of 3.4% of predicted mortality, and her frailty score was at class 5.

The case was discussed in a multidisciplinary Heart Team meeting, and due to the patient's persistent symptoms and recurring hospitalisations, despite the low calcium score, an invasive strategy was decided. The patient was strongly opposed to cardiac surgery, despite her relatively low surgical risk, which contributed to the decision to perform a percutaneous aortic valve implantation.

The patient underwent a transfemoral implantation of a biological aortic prosthesis type *Edwards Sapien 3* of 23 mm, with a good echocardiographic result, mild paravalvular leak, and a mean valve gradient that came down to 8 mmHg.

Immediately after the procedure, a new left bundle branch block was noted (**Figure 1**) that motivated a His-ventricular (HV) exploration. A His-right ventricle conduction delay of 64 ms was found, which increased to 95 ms after Ajmaline provocation. Considering these results and given that the patient was in permanent atrial fibrillation with a difficult-to-control heart rate, despite bitherapy, the decision to implant a pacemaker was made. The patient was implanted with a single-chamber pacemaker SORIN, followed by an atrioventricular node ablation.

One month after the procedure, the patient presented with acute pulmonary oedema. Physical examination revealed a new diastolic heart murmur, and the echocardiography confirmed a seemingly significant paravalvular leak with an ERO (Effective Regurgitant Orifice) by PISA method at 0.1 cm^2 and a planimetry of 0.4 cm^2.

Given the new findings, the patient was referred for percutaneous repair of the paravalvular leak and was successfully implanted with an *Amplatzer Vascular Plug 4* of 8 mm (**Figure 2B**), resulting in a significant reduction of regurgitation and resolution of the heart murmur. Post-procedural echocardiography showed a residual paravalvular leak involving 1/5 of the valve perimeter, a calculated valve surface of 1 cm^2, and a trans-aortic mean gradient of 14 mmHg.

Five weeks after the valvular leak repair, the patient was readmitted for acute heart failure. At this time, echocardiography showed mixed aortic disease with aortic stenosis characterised by a surface of 1 cm^2, a mean gradient of 23 mmHg, and a significant paravalvular aortic regurgitation involving 1/3 of the valve perimeter (**Figure 3**). The case was reconsidered by the Heart Team, and a surgical approach was proposed.

During the surgical procedure, it was found that the *Amplatzer* had moved from its initial position and was oriented towards the leaflets of the bioprosthesis as shown in **Figures 2 A,C**. This contributed to the post-procedural aortic stenosis by impeding the opening of the right coronary leaflet and providing insufficient leak barrier. A surgical aortic valve replacement was

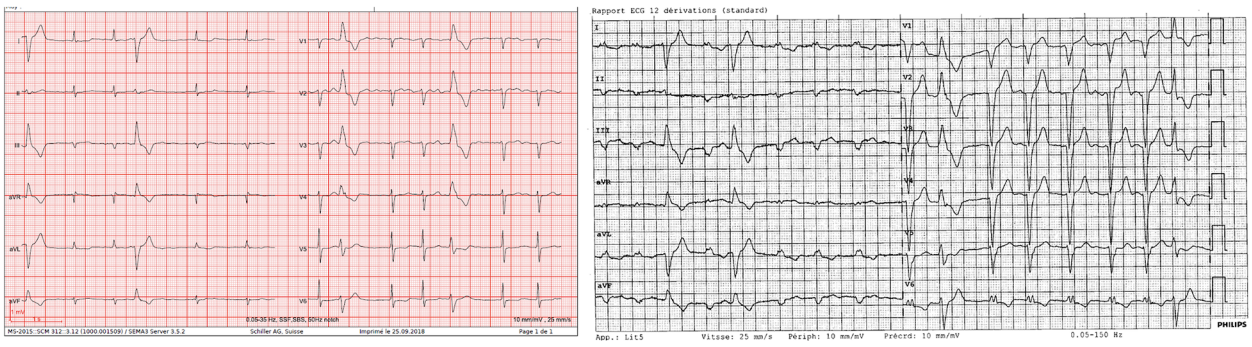

FIGURE 1
Left side: ECG before procedure showing atrial fibrillation, narrow QRS, and two premature ventricular contractions; right side: ECG after procedure showing a large QRS with a left bundle branch block.

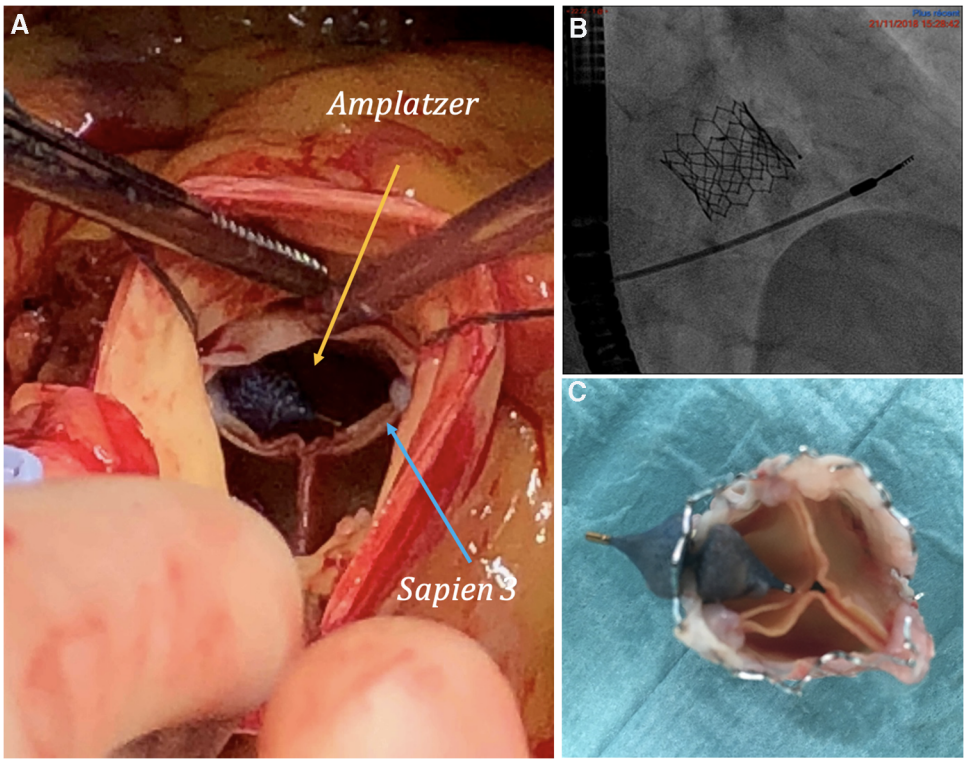

FIGURE 2
(A) Amplatzer vascular plug and bioprosthesis *in vivo*. (B) Angiography of initial implantation of Amplatzer vascular plug. (C) Bioprosthesis *ex vivo* with manual replacement of the Amplatzer vascular plug by the surgeon.

performed using a sutureless LIVANOVA Perceval S bioprosthesis size M, which was completed without any complications. The postoperative echocardiography revealed a preserved left ventricle ejection fraction (62%) with grade 3 diastolic dysfunction, an aortic bioprosthesis with a calculated surface of 0.9 cm^2, and a mean gradient of 18 mmHg with no regurgitation. The patient evolved well and was transferred to cardiac rehabilitation. The mid-term follow-up at 4 years showed a benefit of the procedures in terms of symptoms, with the patient now being in NYHA class I, and hospitalisations, with only one hospitalisation for acute heart failure since being discharged.

Discussion

This case highlights two common complications of TAVI procedures and, more importantly, a complication of percutaneous paravalvular leak repair.

In recent years, new data have emerged regarding electrical conduction complications, when comparing surgical to percutaneous aortic valve replacement. For instance, the incidence of a new left bundle branch block is found to be 10.5%–34.3% in TAVI procedures (7), compared to 4% in surgical aortic valve replacements (8). Additionally, 7.3% of the patients undergoing percutaneous aortic valve replacement end up needing a permanent pacemaker implantation (all causes combined), compared to 3.4% in the surgical series ($p = 0.014$) (9). New left bundle branch block and new pacemaker implantation are some of the few outcomes that favour surgical approach.

With regards to the mechanical complication, this patient initially presented with a mild paravalvular leak that later progressed to a more significant one. The mechanism of this progression is unknown to date. Progression of paravalvular leaks has been described by the PARTNER trial (10); however, it happened over years instead of months seen in our case. So far, no mechanism has been proposed to explain the improvement or worsening of PVL, and measurement methods may explain, in part, these findings. The calcium volume described in the CT scan was low, and there were no indicators of preferential deposition in the device landing zone that could help predict this outcome.

Percutaneous valvular leak repair is a first-line treatment for paravalvular leaks and is nowadays associated with high success rates and few serious complications (11). However, it is a delicate and complex procedure and the correct choice of device and placement is essential. The literature concerning percutaneous paravalvular leak closure after TAVI is relatively scarce, and only a few case series have been published. The number of closures is too low to provide a good understanding of the complications. In contrast, more cases of complications after percutaneous PVL closure have been described in surgical series, and a few larger studies have been published in the literature that describe the most frequent complications encountered (**Table 1**).

TABLE 1 Literature review of case series of Percutaneous PVL closures and its complications.

First author	Year	PVL closures (*n*)	Type of procedure	Number and type of complications
Arri (12)	2015	5	TAVI	1 patient (20%) needed a second device implantation due to complexity of the leak
Gérardin (13)	2019	7	TAVI	No complications
Sorajja (14)	2011	154	Aortic 21%, mitral 79%	11 patients (8.7%) due to either inability to cross the defect, prosthetic leaflet impingement from the occluder, or persistent severe regurgitation
			Bioprosthesis 39%, mechanical 61%	
Ruiz (15)	2011	49	Aortic 22%, mitral 78%	8 patients (16%) due to inability to cross the defect with the delivery system, 3 patients (6%) due to device interference with the mechanical function of the valve prosthesis, and 1 patient (2%) due to wire entrapment during the attempt to cross the aortic paravalvular leak
			Bioprosthesis 65%, mechanical 35%	
Sorajja (16)	2011	115	Aortic 22%, mitral 78%	5 patients (4%) due to prosthetic leaflet impingement, 2 patients (1.7%) due to inability to cross with delivery sheath, 1 patient (0.9%) due to inability to cross with guidewire, 18 patients (15.6%) due to device deployed with residual moderate or severe regurgitation, and 1 patient (0.9%) due to malposition in left ventricle (LV)
			Bioprosthesis 37%, mechanical 63%	

TAVI, transcatheter aortic valve implantation.

In our patient's case, the Amplatzer device appears to have moved and intertwined with the metallic stenting of the prosthesis, which not only caused a failure in completely resolving the leak but also restricted the valve's full opening and led to a reduction in the valve surface, resulting in a new aortic stenosis. The reason for the device's migration is unknown, but it could be related to its initial position during the procedure.

These complications highlight the ongoing debate on indications between TAVI and surgical approaches. Despite the patient's vehement objection to a surgical approach, in light of

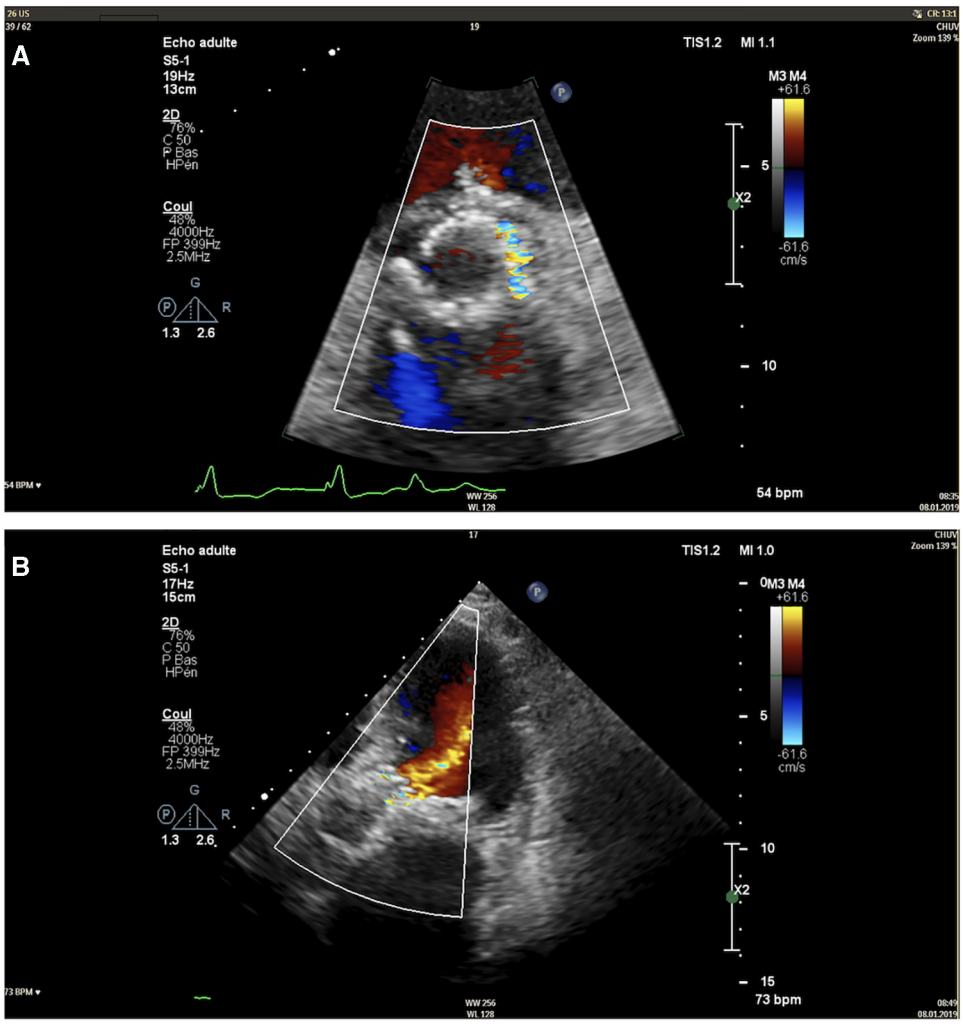

FIGURE 3
Transthoracic echocardiography image of the paravalvular leak in short-axis view (A) and apical five-chamber view (B).

her Euroscore/STS score and frailty score, and considering the small size of the valve annulus, a surgical approach may have yielded better results in this case. Ultimately, the patient underwent surgery anyway.

Conclusion

To the best of our knowledge, this is the first case in which a TAVI paravalvular leak treated percutaneously with an *Amplatzer* device was complicated by the device's migration through the stenting of the bioprosthesis, resulting in a new symptomatic stenosis that required surgery. This highlights the need for better criteria for selecting the appropriate technique for managing paravalvular leaks after TAVI. Whether a baseline surgical approach or valve post-dilation would have been a better option in this patient remains unclear.

Data availability statement

The original contributions presented in the study are included in the article, further inquiries can be directed to the corresponding author.

Ethics statement

Written informed consent was obtained from the next of kin for the publication of any potentially identifiable images or data included in this article.

Author contributions

BPG drafted the manuscript and was responsible for the patient follow-up. OR, EE, and MK provided critical review of the manuscript. EE performed the TAVI and the percutaneous valvular leak repair. MK performed the surgical valve replacement. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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References

1. Vahanian A, Beyersdorf F, Praz F, Milojevic M, Baldus S, Bauersachs J, et al. 2021 ESC/EACTS Guidelines for the management of valvular heart disease. *Eur Heart J*. (2022) 43(7):561–632. doi: 10.1093/eurheartj/ehab395

2. Mack MJ, Leon MB, Thourani VH, Makkar R, Kodali SK, Russo M, et al. Transcatheter aortic-valve replacement with a balloon-expandable valve in low-risk patients. *N Engl J Med*. (2019) 380(18):1695–705. doi: 10.1056/NEJMoa1814052

3. Détaint D, Lepage L, Himbert D, Brochet E, Messika-Zeitoun D, Lung B, et al. Determinants of significant paravalvular regurgitation after transcatheter aortic valve: implantation impact of device and annulus discongruence. *JACC Cardiovasc Interv*. (2009) 2(9):821–7. doi: 10.1016/j.jcin.2009.07.003

4. Leon MB, Smith CR, Mack M, Miller DC, Moses JW, Svensson LG, et al. Transcatheter aortic-valve implantation for aortic stenosis in patients who cannot undergo surgery. *N Engl J Med*. (2010) 363(17):1597–607. doi: 10.1056/NEJMoa1008232

5. Takagi H, Umemoto T. Impact of paravalvular aortic regurgitation after transcatheter aortic valve implantation on survival. *Int J Cardiol*. (2016) 221:46–51. doi: 10.1016/j.ijcard.2016.07.006

6. Athappan G, Patvardhan E, Tuzcu EM, Svensson LG, Lemos PA, Fraccaro C, et al. Incidence, predictors, and outcomes of aortic regurgitation after transcatheter aortic valve replacement: meta-analysis and systematic review of literature. *J Am Coll Cardiol*. (2013) 61(15):1585–95. doi: 10.1016/j.jacc.2013.01.047

7. Roten L, Meier B. Left bundle branch block after transcatheter aortic valve implantation: still a matter of concern? *JACC Cardiovasc Interv*. (2014) 7(2):137–9. doi: 10.1016/j.jcin.2013.11.006

8. Poels TT, Houthuizen P, Garsse LAFMV, Hamad MAS, Maessen JG, Prinzen FW, et al. Frequency and prognosis of new bundle branch block induced by surgical aortic valve replacement. *Eur J Cardiothorac Surg*. (2015) 47(2):e47–53. doi: 10.1093/ejcts/ezu435

9. Bagur R, Rodés-Cabau J, Gurvitch R, Dumont É, Velianou JL, Manazzoni J, et al. Need for permanent pacemaker as a complication of transcatheter aortic valve implantation and surgical aortic valve replacement in elderly patients with severe aortic stenosis and similar baseline electrocardiographic findings. *JACC Cardiovasc Interv*. (2012) 5(5):540–51. doi: 10.1016/j.jcin.2012.03.004

10. Kodali SK, Williams MR, Smith CR, Svensson LG, Webb JG, Makkar RR, et al. Two-year outcomes after transcatheter or surgical aortic-valve replacement. *N Engl J Med*. (2012) 366(18):1686–95. doi: 10.1056/NEJMoa1200384

11. Sorajja P, et al. Percutaneous repair of paravalvular prosthetic regurgitation: patient selection, techniques and outcomes. *Heart*. (2015) 101(9):665–73. doi: 10.1136/heartjnl-2014-306270

12. Arri SS, Poliacikova P, Hildick-Smith D. Percutaneous paravalvular leak closure for symptomatic aortic regurgitation after CoreValve transcatheter aortic valve implantation. *Catheter Cardiovasc Interv*. (2015) 85(4):657–64. doi: 10.1002/ccd.25730

13. Gérardin B, Champagnac D, Smolka G, Bouvaist H, Jakamy R, Ghostine S, et al. Fermeture des fuites para-prothétiques des TAVI. *Ann Cardiol D'Angéiol*. (2019) 68(6):453–61. doi: 10.1016/j.ancard.2019.09.027

14. Sorajja P, et al. Long-term follow-up of percutaneous repair of paravalvular prosthetic regurgitation. *J Am Coll Cardiol*. (2011) 58(21):2218–24. doi: 10.1016/j.jacc.2011.07.041

15. Ruiz CE, Jelnin V, Kronzon I, Dudiy Y, Valle-Fernandez RD, Einhorn BN, et al. Clinical outcomes in patients undergoing percutaneous closure of periprosthetic paravalvular leaks. *J Am Coll Cardiol*. (2011) 58(21):2210–7. doi: 10.1016/j.jacc.2011.03.074

16. Sorajja P, et al. Percutaneous repair of paravalvular prosthetic regurgitation. *Circulation*. (2011) 4(4):314–21. doi: 10.1161/CIRCINTERVENTIONS.110.960955

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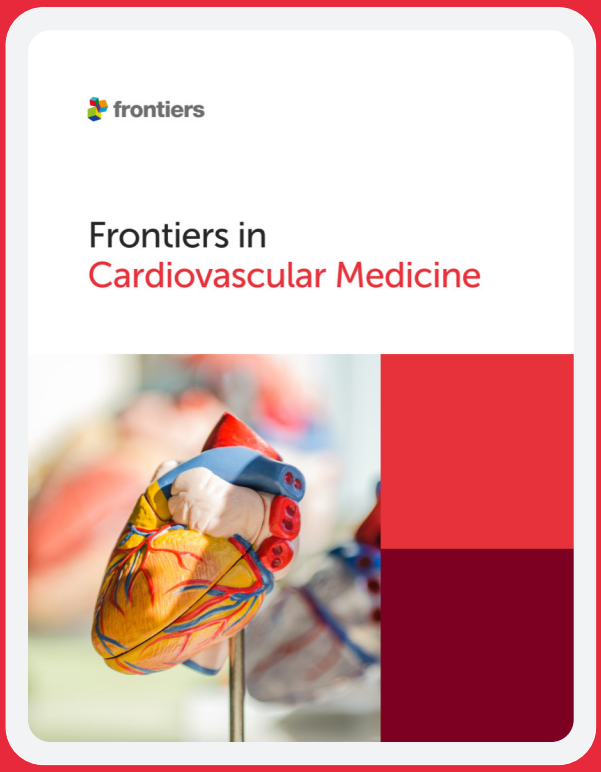

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