frontiers | Research Topics

Case reports in heart surgery 2024

Edited by
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Published in
Frontiers in Cardiovascular Medicine

ISSN 1664-8714
ISBN 978-2-8325-7125-5
DOI 10.3389/978-2-8325-7125-5

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Case reports in heart surgery: 2024

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Citation
Gatti, G., ed. (2025). *Case reports in heart surgery: 2024*.
Lausanne: Frontiers Media SA. doi: 10.3389/978-2-8325-7125-5

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frontiers | Frontiers in Cardiovascular Medicine

TYPE Editorial
PUBLISHED 29 October 2025
DOI 10.3389/fcvm.2025.1692834

OPEN ACCESS

EDITED AND REVIEWED BY
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RECEIVED 26 August 2025
ACCEPTED 07 October 2025
PUBLISHED 29 October 2025

CITATION
Gatti G (2025) Editorial: Case reports in heart surgery 2024.
Front. Cardiovasc. Med. 12:1692834.
doi: 10.3389/fcvm.2025.1692834

Editorial: Case reports in heart surgery 2024

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KEYWORDS

left ventricular aneurysm, left ventricular thrombosis, mediastinitis, multidisciplinary approaches, multimodal imaging, case report, heart surgery

Editorial on the Research Topic
Case reports in heart surgery: 2024

For the second consecutive year, I had the pleasure and honor of coordinating the (fourth) Research Topic of "*Case Reports in Heart Surgery*" by Frontiers in Cardiovascular Medicine and Frontiers in Surgery. I am sincerely grateful to the Editors-in-Chief for their renewed trust.

As with the previous edition, the objective for the 2024 Research Topic was to feature unique cases of patients that present with an unexpected diagnosis, treatment outcomes, or clinical courses. Only original Case Reports that will significantly advance the field of cardiac surgery were considered, in my opinion and that of the Reviewers who collaborated with me throughout the manuscript review process. Rare cases with typical features, frequent cases with atypical features, and cases with a convincing response to new treatments were included in the present Research Topic.

The (undeclared) goal was also to match the excellent results achieved by the 2023 edition of the Research Topic, which has received over 26,000 views to date. I am confident! The contributions selected in this 2024 edition are also rare and original clinical cases that could offer readers of Frontiers many points of interest and reflection.

The Research Topic consists of 15 articles written by a total of 80 Authors from seven countries (China, Colombia, Russia, Singapore, Switzerland, Türkiye, and the United States) spanning four different continents. To date, the Research Topic has already received over 17,000 views.

The main issues addressed in the present Research Topic are

- The growing importance of minimally invasive surgery and interventional techniques and technologies (Tian et al., Peng et al., Idu et al., Çetinarslan and Saba, Bidovecet al., Xia et al., Aigumov et al., Chen et al.), and, inevitably, of their complications (Tian et al., Peng et al., Idu et al.). Obviously, the "minimally invasive" concept should be understood in a broad sense, including minimal, mechanical, and/or biological (humoral and cellular) invasiveness;
- The essential need for multimodal imaging for complex cardiovascular lesions (Tian et al., Lianget al., Shan et al., Peng et al., Idu et al., Bidovecet al., Xia et al., Yang et al., Aigumov et al., Chen et al., Ma et al.);
- The essential need for a multidisciplinary approach to diseases involving multiple organs or systems, including the heart (Wang et al., Lianget al., Peng et al.,

TABLE 1 Case reports in heart surgery 2024[a].

Paper number	First author	Title	Publication date	DOI	Total views[b]	Main findings	Keywords
1.	Yunfei Tian	Case Report: Minimally invasive management of suspected active bleeding from intercostal vessel after axillary thoracotomy ventricular septal defect repair: an application of the Foley catheter	May 27, 2025	doi.org/10.3389/fcvm.2025.1511221	640	Injury to the intercostal vessels during an axillary thoracotomy can be a serious complication of minimally invasive cardiac surgery. The Authors reported a case of major bleeding following right axillary thoracotomy that was successfully treated using a Foley catheter inserted through the chest wall.	Axillary thoracotomy; Foley catheter; Minimally invasive surgery
2.	Zhaohui Wang	Case Report: Deep sternal wound infection caused by *Mycoplasma hominis* after cardiac surgery	May 12, 2025	doi.org/10.3389/fcvm.2025.1538389	759	The Authors reported a case of DSWI after cardiac surgery caused by *Mycoplasma hominis*.	DSWI; *Mycoplasma hominis*
3.	Chunshui Liang	Successful treatment of massive biventricular thrombi associated with myocarditis: a case report	Apr 28, 2025	doi.org/10.3389/fcvm.2025.1530548	797	In a young man with a stroke, massive bi-ventricular thrombosis associated with myocarditis was successfully treated with drugs alone, according to a multidisciplinary approach.	Massive bi-ventricular thrombosis; Multidisciplinary approach; Myocarditis
4.	Jianggui Shan	Case Report: "Dumbbell" giant right coronary artery ectasia with right atrial fistula	Feb 28, 2025	doi.org/10.3389/fcvm.2025.1498359	943	Type IV giant ectasia of the right coronary artery, right atrial fistula, epicardial muscle bridge, and functional mitral regurgitation were all successfully repaired in a middle-aged woman. The Authors discussed the origin and developing stages of this complex cardiac lesion.	Cardiac fistula; Giant coronary aneurysm
5.	Danping Peng	Aortic endograft infection after thoracic endovascular aortic repair: two case reports and literature review	Feb 25, 2025	doi.org/10.3389/fcvm.2025.1549613	919	Two unresolved cases of late aortic endograft infection complicated by peri-aortic abscess involving the posterior mediastinum were presented by the Authors, who emphasized the role of strict follow-up after TEVAR to prevent late life-threatening complications. A literature review of the topic was also reported.	Mediastinitis; Peri-aortic abscess; TEVAR
6.	Nicolas Nunez-Ordonez	Case Report: A usual procedure in an unusual situation: a patient with a rare Ehlers-Danlos/ osteogenesis imperfecta overlap undergoing aortic valve replacement	Feb 24, 2025	doi.org/10.3389/fcvm.2025.1480363	1,071	A case of Ehlers-Danlos/ Osteogenesis Imperfecta Overlap Syndrome undergoing aortic valve replacement for aortic regurgitation was presented. The Authors emphasized the importance of a multidisciplinary approach to managing patients with rare genetic disorders.	Ehlers-Danlos syndrome; Multidisciplinary approach; Osteogenesis imperfecta; Overlap syndrome
7.	Muhammad Idu	Case Report: Late presentation of post-coronary artery bypass surgery pericardial effusion heralding mediastinitis with tracheocutaneous fistula	Feb 13, 2025	doi.org/10.3389/fcvm.2025.1483395	1,023	A case of late post-CABG pericardial effusion, complicated by mediastinitis and tracheocutaneous fistula, was illustrated.	Mediastinitis; Pericardial effusion; Tracheocutaneous fistula

(*Continued*)

TABLE 1 Continued

Paper number	First author	Title	Publication date	DOI	Total views[b]	Main findings	Keywords
8.	Özge Çetinarslan	Case Report: Staged surgical management in ESRD: off-pump CABG followed by renal transplantation to enhance graft survival	Feb 7, 2025	doi.org/10.3389/fcvm.2025.1486771	798	Two patients scheduled for renal transplantation because of end-stage renal failure underwent uneventful off-pump CABG without blood transfusion. The Authors emphasized the role of off-pump surgery, compared to on-pump surgery, in minimizing blood transfusions, which could eventually increase the risk of graft rejection.	Blood transfusion; Graft rejection; Off-pump CABG; Minimally invasive surgery
9.	Jan Bidovec	Case Report: PCI of the left coronary artery after salvage operation in a comatose patient with an acute type A aortic dissection	Feb 6, 2025	doi.org/10.3389/fcvm.2025.1516152	1,128	A two-stage, hybrid treatment involving ascending aorta plus aortic arch replacement, along with intravascular ultrasound-guided stenting of the left main, was performed on a hemiplegic elderly woman with bilateral carotid artery and right vertebral artery occlusion owing to acute aortic dissection. Since there was no residual neurological deficit, the Authors highlighted the importance of a multidisciplinary approach.	Acute aortic dissection; Hybrid treatment; Multidisciplinary approach; Neurological deficit
10.	Jianming Xia	Case Report: Surgery combined with extracorporeal membrane oxygenation for acute type A aortic dissection complicated with acute myocardial infarction	Jan 31, 2025	doi.org/10.3389/fcvm.2025.1463764	865	ECMO was successfully used as a perioperative hemodynamic support measure for three patients with acute aortic dissection complicated by right ventricular myocardial infarction.	Acute aortic dissection; ECMO; Peri-operative management
11.	Jeffrey Rodgers	Case Report: *Fusarium falciforme* pericardial and sternal wound infection following orthotopic heart transplantation	Jan 6, 2025	doi.org/10.3389/fcvm.2024.1480392	768	The Authors reported on the successful treatment of *Fusarium falciforme* mediastinitis in a heart transplant recipient.	*Fusarium falciforme*; Heart transplantation; Mediastinitis
12.	Yang Yuehang	Aortic valve replacement in a bicuspid aortic valve patient followed by reoperation for ascending aorta rupture: a case report	Dec 11, 2024	doi.org/10.3389/fcvm.2024.1471686	889	One case of ascending aortic rupture and aortic-right atrial fistula occurred after a replacement of stenotic BAV combined with ascending aortoplasty for mild (43 mm) aortic dilation. Replacement of the ascending aorta was finally performed. Given the increased risk of severe future complications, the Authors raised awareness among the readers about the need to consider a strategy for resolving ascending aortic replacement in the presence of diseased BAV, even with aortic dilation cutoffs lower than 45 mm (as recommended by the most recent guidelines recommendations).	Ascending aortic replacement; Ascending aortic rupture; Ascending aortoplasty; BAV; Guidelines

(*Continued*)

TABLE 1 Continued

Paper number	First author	Title	Publication date	DOI	Total views[b]	Main findings	Keywords
13.	RN Aigumov	Geometric reconstruction of the left ventricle on a beating heart through a minimally invasive approach from the left anterolateral thoracotomy: case report	Dec 4, 2024	doi.org/10.3389/fcvm.2024.1507222	1,043	A post-infarct aneurysm of the LV, complicated by endocavitary thrombosis, was successfully repaired through left anterolateral thoracotomy using the "beating heart" technique. According to the Authors, this minimally invasive approach is safe and could improve LV geometric reconstruction.	Beating heart surgery; LV aneurysm; Minimally invasive surgery
14	Yu Chen	Case Report: 3D imaging-assisted minimally invasive hybrid closure surgery of a complex coronary artery fistula	Nov 22, 2024	doi.org/10.3389/fcvm.2024.1439263	803	A pediatric case of successful, single-stage hybrid closure of a complex coronary artery fistula was reported. The Authors emphasized the decisive role of preoperative three-dimensional imaging and intraoperative transesophageal echocardiography.	Complex coronary artery fistula; Hybrid treatment; Minimally invasive surgery; Three-dimensional imaging
15	Hongjia Ma	Case Report: Surgical management of traumatic giant coronary artery pseudoaneurysm with pericardial patch repair and ostium isolation	Nov 8, 2024	doi.org/10.3389/fcvm.2024.1462557	633	A complex surgical repair of a giant coronary artery pseudoaneurysm caused by trauma was reported. The correct anatomical reconstruction was confirmed with transthoracic echocardiography at one month after surgery.	Cardiac trauma; Giant coronary pseudoaneurysm

BAV, bicuspid aortic valve; CABG, coronary artery bypass grafting; DOI, digital object identifier; DSWI, deep sternal wound infection; ECMO, extracorporeal membrane oxygenator; LV, left ventricle/ventricular; TEVAR, thoracic endovascular aortic repair.
[a]Case reports are listed in order of publication date.
[b]22 Aug, 2025.

Nunez-Ordonez et al., Idu et al., Çetinarslan and Saba, Bidovecet al., Xia et al., Rodgers et al., Yang et al., Chen et al., Ma et al.). To improve patient outcomes, in addition to the cardiologist and cardiac surgeon, other healthcare professionals, each within their own area of expertise, should also be involved in the management of complex cardiac patients and/or diseases. This is in accordance with the Latin maxim, "Unicuique suum" (To each his/her own);

- The significant role of sometimes neglected (albeit life-threatening) complications such as sternal wound infections (Wang et al., Idu et al., Rodgers et al.) and pericardial effusion (Idu et al.);

An interesting report on two unresolved cases of late aortic endograft infection complicated by peri-aortic abscess (Peng et al.) was (perhaps improperly) included in the present Research Topic of case reports on heart surgery to familiarize cardiac surgeons with endovascular repair of the thoracic aorta and its rare but catastrophic complications involving the mediastinum.

I synthesized the main message of each contribution to the present Research Topic in Table 1.

In the 2023 Editorial (1), I stated: "Personally, I am particularly fond of the Case Reports sections of surgical Journals because they often include interesting and innovative contributions. The clinical presentation, diagnostic process and effective surgical treatment of rare conditions offer the reader stimulating food for thought. Sometimes there are reported cases of failure but of great educational value. However, Case Reports sections are increasingly rare nowadays in scientific Journals where more value is placed on large-scale studies such as multicenter studies, randomized controlled trials or meta-analyses. (..) Both for Heart Surgery and Interventional Cardiology, the most advanced frontiers of the disciplines are often glimpsed by analyzing Case Reports!". [SIC] Today, after reviewing dozens of Case Reports for Frontiers, I agree with myself more than ever.

In closing, I would like to sincerely thank all the valuable Reviewers and Co-editors who helped me with my task. I have certainly learned a great deal from them throughout this experience. In addition, I would like to thank all the members of the Editorial Offices of the two valuable scientific Journals for supporting me at every step of this experience. Thank you again to everyone!

Author contributions

GG: Formal analysis, Visualization, Investigation, Data curation, Resources, Validation, Software, Writing – review & editing, Writing – original draft, Methodology, Conceptualization.

Funding

The author(s) declare that no financial support was received for the research, and/or publication of this article.

Conflict of interest

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

The author(s) declared that they were an editorial board member of Frontiers, at the time of submission. This had no impact on the peer review process and the final decision.

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Publisher's note

References

1. Gatti G. Editorial: case reports in heart surgery: 2023. *Front Cardiovasc Med*. (2024) 11:1527906. doi: 10.3389/fcvm.2024.1527906

frontiers | Frontiers in Cardiovascular Medicine

TYPE Case Report
PUBLISHED 08 November 2024
DOI 10.3389/fcvm.2024.1462557

OPEN ACCESS

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RECEIVED 10 July 2024
ACCEPTED 16 October 2024
PUBLISHED 08 November 2024

CITATION
Ma H, Qian H and Meng W (2024) Case Report: Surgical management of traumatic giant coronary artery pseudoaneurysm with pericardial patch repair and ostium isolation. Front. Cardiovasc. Med. 11:1462557. doi: 10.3389/fcvm.2024.1462557

Case Report: Surgical management of traumatic giant coronary artery pseudoaneurysm with pericardial patch repair and ostium isolation

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There is limited literature regarding cases of giant coronary artery aneurysms (GCAAs), and instances of giant coronary artery pseudoaneurysms caused by trauma are exceedingly rare. Here is a case presentation of an adult male who developed a giant coronary artery pseudoaneurysm following trauma. Successful surgical intervention was performed, involving repair of the aneurysmal opening with a pericardial patch and isolation of the right coronary artery ostium into the aortic root. One month postoperatively, a follow-up transthoracic echocardiogram revealed thrombotic occlusion within the residual lumen of the right coronary artery aneurysm, with contiguous echogenicity extending from the aortic sinus to the right coronary artery.

KEYWORDS

coronary artery aneurysm, coronary artery pseudoaneurysm, giant coronary artery pseudoaneurysm, surgical intervention, trauma

Introduction

The notion of coronary artery aneurysm was originally postulated by Morgagni in 1761 (1). Presently, it is delineated as an expansion of the vascular structure with a diameter ≥1.5 times that of the adjacent normative vessel. Infrequently, this dilation may reach dimensions warranting classification as a giant coronary artery aneurysm, a concept subject to ongoing debate concerning precise criteria. Diverse definitions have been proposed by scholars, delineating aneurysms exceeding diameters of 20 mm, 40 mm, or 50 mm, or those surpassing fourfold the reference vessel diameter (2). Literature suggests that merely 0.02% of coronary artery aneurysms meet the stringent criteria for classification as giant coronary artery aneurysms (3, 4). The etiology of these aneurysms is multifaceted, with atherosclerosis emerging as the predominant cause, Coronary artery pseudoaneurysms are rare and often occur after catheter-based interventions, surgical complications, blunt trauma, or infection (5). Herein, we present a case involving an adult male patient diagnosed with a giant coronary artery pseudoaneurysm subsequent to trauma, who underwent successful surgical intervention.

Case presentation

The patient is a 54-year-old male who experienced chest pain after colliding with a car while riding a motorcycle, resulting in a 16-h coma before presenting to our emergency

department. There was no primary loss of consciousness after the injury, no evidence of open chest trauma, and no respiratory distress. He denies any family history of genetic diseases or past cardiac history, previous transthoracic echocardiography did not mention any abnormalities. Following initial treatment at a local hospital, his condition deteriorated gradually, leading to altered mental status, prompting endotracheal intubation and assisted ventilation with a mechanical ventilator.

Upon admission, vital signs were stable, and a computed tomography scan of the head revealed no significant abnormalities. Left ventricular ejection fraction was measured at 71%. The preoperative electrocardiogram indicated mild ST-T segment depression in the interventricular septum and lateral wall, with a peak troponin-T level of 872.0 ng/L.

Transthoracic echocardiography (Figure 1) revealed a tumor-like abnormal hypoechoic area measuring approximately 70 mm × 66 mm × 75 mm located about 1 cm from the aortic valve annulus, extending outward from the sinus. This mass communicated with the aorta through a 15 mm × 9 mm defect, with blood flow directed from the aorta to the right coronary artery. Contrast-enhanced computed tomography angiography (Figure 2) confirmed a spherical dilatation (6.0 cm × 7.5 cm) in the right anterior aspect of the ascending aorta. A defect measuring approximately 1.1 cm in diameter was seen in the anterior aspect of the ascending aorta, connecting with the spherical dilation. Compression was noted on the ascending aorta root, aortic sinuses, and right atrium.

Following meticulous deliberation, surgical intervention is deemed necessary considering the risk of aneurysm rupture. The procedure was conducted through a median sternotomy, meticulously achieving hemostasis in a stepwise manner, followed by a gradual opening of the pericardium, revealing an accumulation of approximately 100 mL of aged, dark red hemopericardium, concomitant with partial thrombus formation

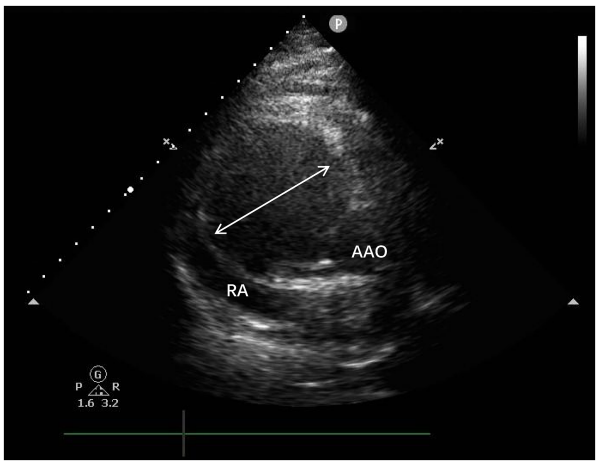

FIGURE 1
A tumor-like abnormal hypoechoic area (white arrow) measuring approximately 70 mm × 66 mm × 75 mm located about 1 cm from the aortic valve annulus, extending outward from the sinus. RA, right atrium; AAO, ascending aortat.

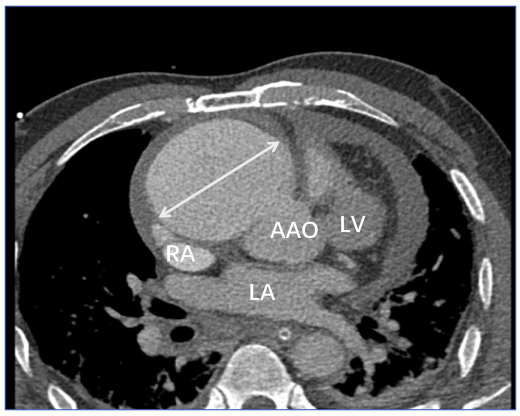

FIGURE 2
Contrast-enhanced computed tomography angiography confirmed a spherical dilatation (6.0 cm × 7.5 cm) in the right anterior aspect of the ascending aorta (white arrow). LA, left atrium; LV, left ventricle; RA, right atrium; AAO, ascending aorta.

(Figure 3). Remarkably, a substantial aneurysm was discerned at the aortic root and atrioventricular groove, measuring approximately 12 cm × 10 cm (Figure 4). Establishment of cardiopulmonary bypass ensued, achieved through cannulation of the ascending aorta and femoral vein. Following cardiac arrest, the aneurysm was meticulously incised for exploration, unveiling a 1.5 cm × 2 cm communication between the right coronary sinus wall and the aneurysm, with the right coronary artery ostium positioned 5 mm proximal to the opening (Figure 5). Intraoperatively, in light of the operative perils entailed in aneurysm extirpation and aortic root reconstruction, and judiciously considering the prospective postoperative outcomes, it was decided to employ a 2.5 cm × 2.5 cm pericardial patch to wrap around the openings of the aneurysm at two sites, forming a conduit between them, facilitating the isolation of the remaining portion of the aneurysm while maintaining coronary blood flow (Figure 6). Subsequent to aortotomy, no notable hemorrhage emanated from the patch. Intraoperative transesophageal echocardiography confirmed the cessation of the aortic-to-aneurysm shunt. Successful weaning from cardiopulmonary bypass ensued, and thoracic closure ensued in accordance with established protocols. The duration of cardiopulmonary bypass was 67 min. Postoperative histopathological examination delineated fibrous tissue hyperplasia, hyalinization, and thrombus deposition, consistent with alterations characteristic of the "aneurysmal wall" (Figure 7).

One month postoperative, the patient attended a follow-up appointment at our institution. Computed tomography angiography (CTA) revealed localized nodular protrusions in the right coronary sinus, measuring approximately 1.4 cm × 1.2 cm (Figure 8). Transthoracic echocardiography (TTE) demonstrated continuous echogenicity along the wall of the aortic sinus to the right coronary artery, indicative of thrombosis within the residual lumen of the right coronary artery aneurysm (Figure 9).

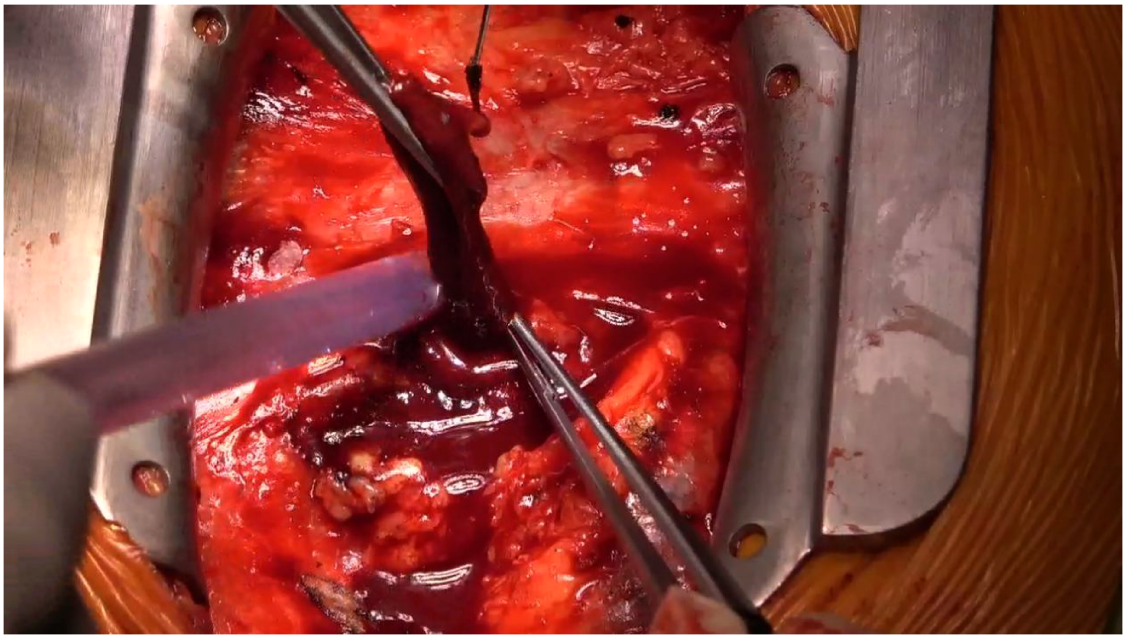

FIGURE 3
Within the pericardium, there is approximately 100 mL of dark red, old blood clots, along with partial thrombus formation.

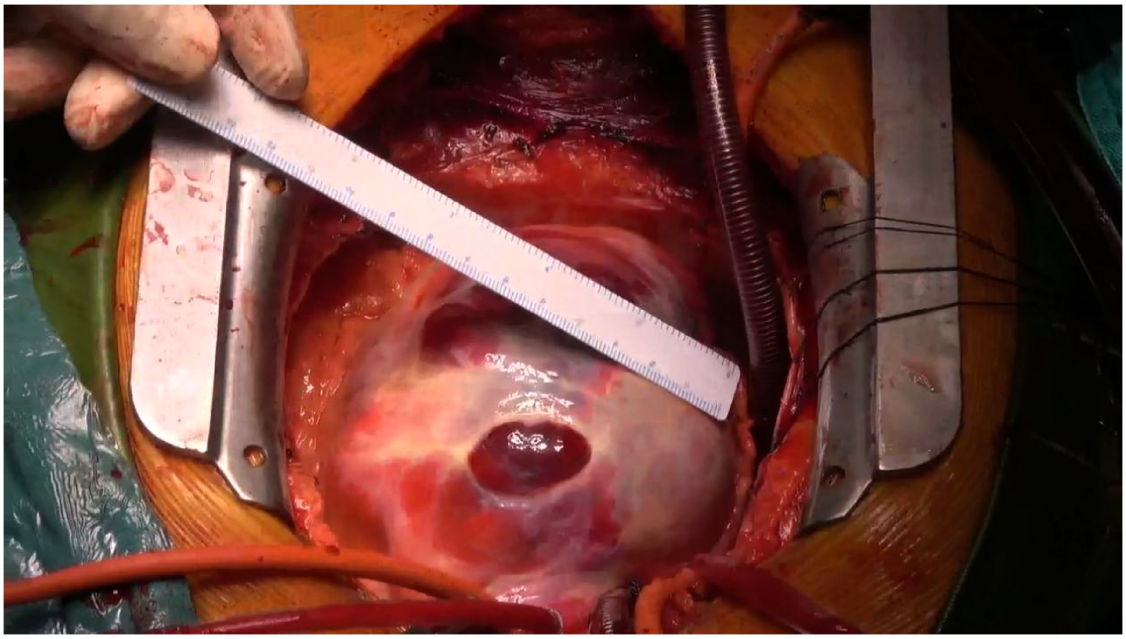

FIGURE 4
In the aortic root and atrioventricular groove, there is evident presence of a substantial mass, measuring approximately 12 cm × 10 cm.

Discussion

Coronary artery aneurysms (CAAs) can often remain asymptomatic, yet their symptomatic manifestation frequently involves acute coronary syndrome or respiratory distress (6). Conversely, GCAAs demonstrate diverse and occasionally ambiguous clinical presentations, occasionally resembling mediastinal masses or cardiac neoplasms (7).

In this case, the patient's clinical course was brief, and the CT scan indicated a low-density fluid area surrounding the contrast-enhanced region. Pathological analysis further revealed fibrous hyperplasia, hyaline degeneration of the vessel wall, and

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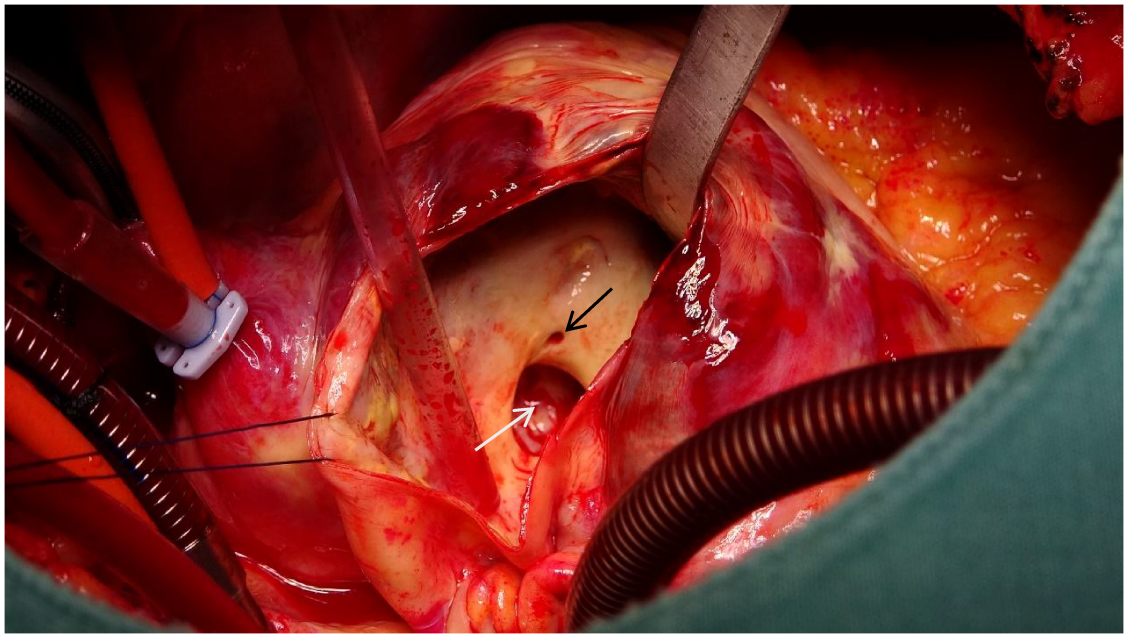

FIGURE 5
A 1.5 cm × 2 cm communication between the right coronary sinus wall and the aneurysm (white arrow), with the right coronary artery ostium (black arrow) positioned 5 mm proximal to the opening.

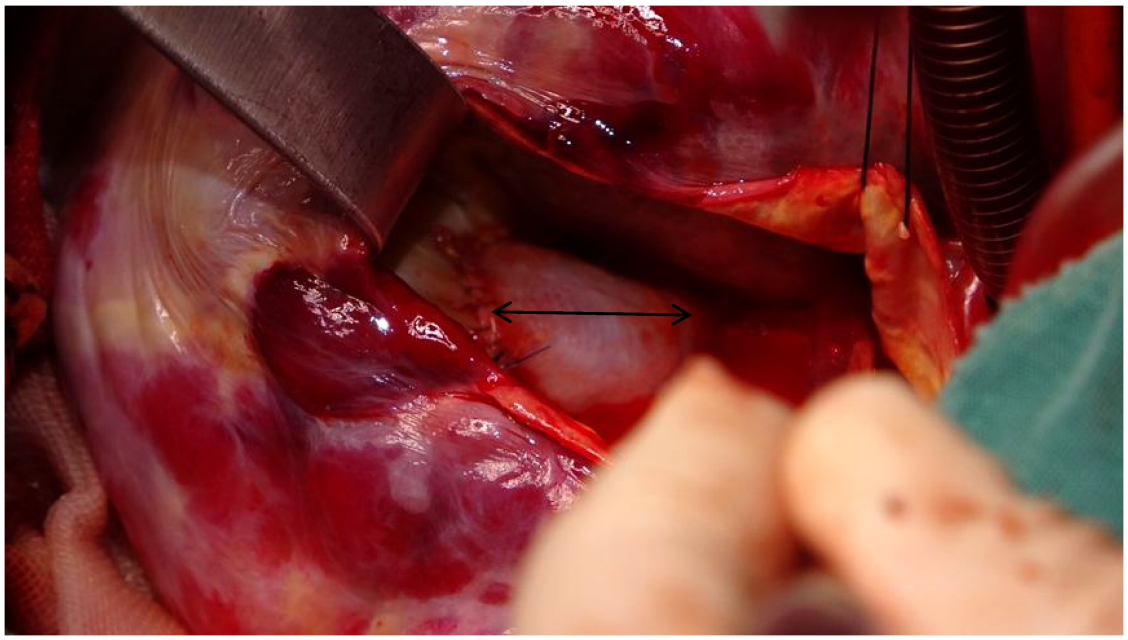

FIGURE 6
Utilizing a 2.5 cm × 2 cm pericardial patch (black arrow) to close the passage between the aortic root and the aneurysm, while resecting the ostium of the right coronary artery into the aortic root.

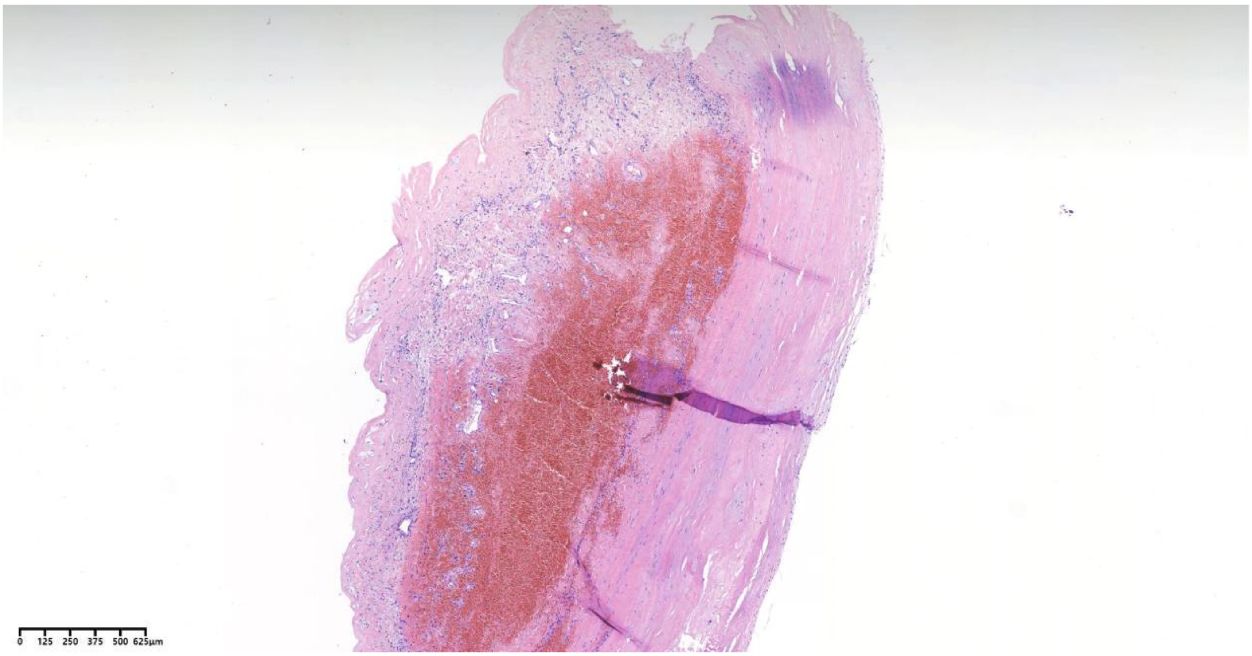

FIGURE 7
Pathological analysis employing hematoxylin-eosin staining reveals vascular wall fibrous hyperplasia, hyaline degeneration, and thrombus formation.

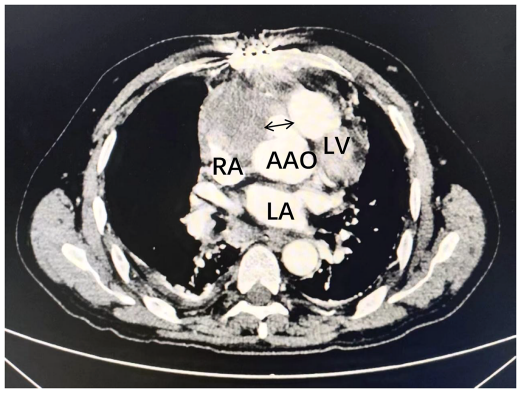

FIGURE 8
Computed tomography angiography (CTA) suggests localized nodular protrusions in the right coronary sinus, measuring approximately 1.4 × 1.2 cm (black arrow). LA, left atrium; LV, left ventricle; RA, right atrium; AAO, ascending aorta.

FIGURE 9
Transthoracic echocardiography (TTE) demonstrated continuous echogenicity along the wall of the aortic sinus to the right coronary artery, indicative of thrombosis within the residual lumen (white arrow) of the right coronary artery aneurysm.

thrombus formation. Consequently, we postulate that the patient developed a giant coronary artery pseudoaneurysm as a result of the traumatic injury (8).

In cases of coronary artery pseudoaneurysms, patient presentations can vary widely, ranging from asymptomatic to myocardial infarction, ventricular arrhythmias, or respiratory symptoms (9). In this report, the rapid formation of a coronary artery pseudoaneurysm is considered to have led to cardiac compression, resulting in transient cardiac insufficiency accompanied by loss of consciousness.

The management of CAAs should be individualized based on the patient's unique clinical presentation. When underlying primary diseases are present, therapeutic strategies should primarily address these underlying conditions. Specific treatment modalities for CAAs encompass both interventional and surgical interventions, necessitating a thorough evaluation of factors including aneurysm dimensions, anatomical location, and etiological factors (10). The treatment approach for coronary artery pseudoaneurysms is similar to that for coronary artery aneurysms, but it requires additional consideration of the heightened risk of pseudoaneurysm rupture (11, 12). In the

context of this case report, the proximity of the coronary artery ostium to the aneurysmal opening raises concerns regarding potential obstruction of the coronary artery ostium with interventional embolization. Consequently, surgical exploration and repair of the opening are deemed appropriate and efficacious.

Conclusion

Giant coronary artery pseudoaneurysms are relatively rare and have diverse etiologies. Therefore, treatment for giant coronary artery pseudoaneurysms needs to be individualized based on the patient's condition, addressing both the underlying primary disease and the aneurysm itself. Currently, there is a lack of sufficient evidence to support specific treatment strategies for post-traumatic giant coronary artery pseudoaneurysms, highlighting the need for further research.

Data availability statement

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

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

HM: Writing – original draft. HQ: Writing – review & editing. WM: Writing – original draft, Writing – review & editing.

Funding

The author(s) declare that no financial support was received for the research, authorship, and/or publication of this article.

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

References

1. Pham V, Hemptinne Q, Grinda JM, Duboc D, Varenne O, Picard F. Giant coronary aneurysms, from diagnosis to treatment: a literature review. *Arch Cardiovasc Dis*. (2020) 113(1):59–69. doi: 10.1016/j.acvd.2019.10.008

2. Eshtehardi P, Cook S, Moarof I, Triller HJ, Windecker S. Giant coronary artery aneurysm: imaging findings before and after treatment with a polytetrafluoroethylene-covered stent. *Circ Cardiovasc Interv*. (2008) 1(1):85–6. doi: 10.1161/CIRCINTERVENTIONS.107.763656

3. Ko K, Kroeze V, Heijmen RH, Verkroost M, Smith T. Surgical treatment of a giant right coronary aneurysm. *Multimed Man Cardiothorac Surg*. (2024) 2024. doi: 10.1510/mmcts.2023.099

4. Morita H, Ozawa H, Yamazaki S, Yamauchi Y, Tsuji M, Katsumata T, et al. A case of giant coronary artery aneurysm with fistulous connection to the pulmonary artery: a case report and review of the literature. *Intern Med*. (2012) 51(11):1361–6. doi: 10.2169/internalmedicine.51.7134

5. Dhakam S, Ahmed H, Jafferani A. Percutaneous coronary intervention of left main pseudoaneurysm with customized covered stents. *Catheter Cardiovasc Interv*. (2011) 77(7):1033–5. doi: 10.1002/ccd.22903

6. Arboine L, Palacios JM. Left main coronary artery aneurysm. *N Engl J Med*. (2018) 378(23):e32. doi: 10.1056/NEJMicm1708877

7. Dadkhah Tirani H, Aghajanzadeh M, Pourbahador R, Hassanzadeh R, Ebrahimi H. Giant right coronary artery aneurysm mimicking a mediastinal cyst with compression effects: a case report. *Res Cardiovasc Med*. (2016) 5(3):e32086. doi: 10.5812/cardiovascmed.32086

8. Yoshioka D, Izutani H, Ryugo M, Kawachi K, Sawa Y. Asymptomatic giant traumatic right coronary artery pseudoaneurysm caused by sternal fracture. *Ann Thorac Surg*. (2011) 92(2):e33–5. doi: 10.1016/j.athoracsur.2011.03.046

9. Aqel RA, Zoghbi GJ, Iskandrian A. Spontaneous coronary artery dissection, aneurysms, and pseudoaneurysms: a review. *Echocardiography*. (2004) 21(2):175–82. doi: 10.1111/j.0742-2822.2004.03050.x

10. Zeb M, McKenzie DB, Scott PA, Talwar S. Treatment of coronary aneurysms with covered stents: a review with illustrated case. *J Invasive Cardiol*. (2012) 24 (9):465–9.

11. Kar S, Webel RR. Diagnosis and treatment of spontaneous coronary artery pseudoaneurysm: rare anomaly with potentially significant clinical implications. *Catheter Cardiovasc Interv*. (2017) 90(4):589–97. doi: 10.1002/ccd.26997

12. Wang YC, Hsu RB, Huang CH. Spontaneous giant right coronary artery pseudoaneurysm. *J Thorac Cardiovasc Surg*. (2014) 148(1):349–50. doi: 10.1016/j.jtcvs.2013.12.023

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TYPE Case Report
PUBLISHED 22 November 2024
DOI 10.3389/fcvm.2024.1439263

OPEN ACCESS

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RECEIVED 27 May 2024
ACCEPTED 06 November 2024
PUBLISHED 22 November 2024

CITATION
Chen Y, Lu J, Lian X, Chang P, Wen P, Ma L and Liu Y (2024) Case Report: 3D imaging-assisted minimally-invasive hybrid closure surgery of a complex coronary artery fistulas.
Front. Cardiovasc. Med. 11:1439263.
doi: 10.3389/fcvm.2024.1439263

Case Report: 3D imaging-assisted minimally-invasive hybrid closure surgery of a complex coronary artery fistulas

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Coronary artery fistulas (CAFs) are rare congenital heart defects that are typically managed through interventional closure, traditional surgery, or minimally invasive hybrid closure surgery. However, treating CAFs with complex anatomy, such as tortuous vessels, presents a significant challenge, particularly in young children. We report the case of a 3.8-year-old child (15 kg/100 cm) with a complex CAF, treated using a minimally invasive hybrid closure surgery approach with a 4 × 4 mm Amplatzer Duct Occluder II (ADO II) (Abbott, USA). Three-dimensional (3D) imaging was utilized to visualize the CAF's anatomy, guide the surgical planning, and accurately determine the puncture site on the right ventricular free wall, as well as the optimal sheath direction and insertion depth. The procedure was carried out efficiently and safely, guided by preoperative 3D imaging and intraoperative transesophageal echocardiography. Follow-up at one year demonstrated excellent outcomes with no complications.

KEYWORDS

coronary artery fistula, congenital heart disease, three-dimensional imaging, minimally invasive hybrid closure surgery, case report

1 Introduction

Coronary artery fistulas (CAFs), which account for approximately 0.2%–0.4% of all congenital heart diseases (1, 2), are very rare coronary artery malformations, occurring with an incidence of 0.1%–0.22%. They generally do not present with clinical signs or symptoms and are usually discovered incidentally. However, the likelihood of their spontaneous closure is very small, and large and/or symptomatic CAFs have an extremely high chance of developing late symptoms and complications. Therefore, early treatment is necessary once a CAF is diagnosed (3, 4). Treatment methods for CAFs include interventional closure, traditional surgery, and minimally invasive hybrid closure surgery. Traditional surgery is suitable for almost all CAFs, whereas interventional closure is more suited for patients with a more accessible fistula or straight fistula course. However, traditional surgery and interventional closure are more challenging for CAFs that have complex anatomical structures and deep anatomical locations and are difficult to expose. Herein, we report a case of a patient with a CAF who was treated with minimally invasive hybrid closure surgery, which was successfully performed under the guidance of preoperative three-dimensional (3D) imaging and intraoperative transesophageal echocardiography (TEE).

2 Case report

A 2.8-year-old boy was referred to our institution for a grade III/IV systolic heart murmur detected during a physical examination for pneumonia. Echocardiography revealed slight enlargement of the left heart and dilation of the right coronary artery (RCA), with an internal diameter of approximately 5 mm. A 4 mm CAF was observed in the apical part of the right ventricle (RV), with a left-to-right shunt, and a maximal continuous flow velocity of 4.4 m/s. Cardiac computed tomography angiography (CTA) (Figure 1A–C) confirmed that the RCA was dilated giving birth to a CAF extending from the

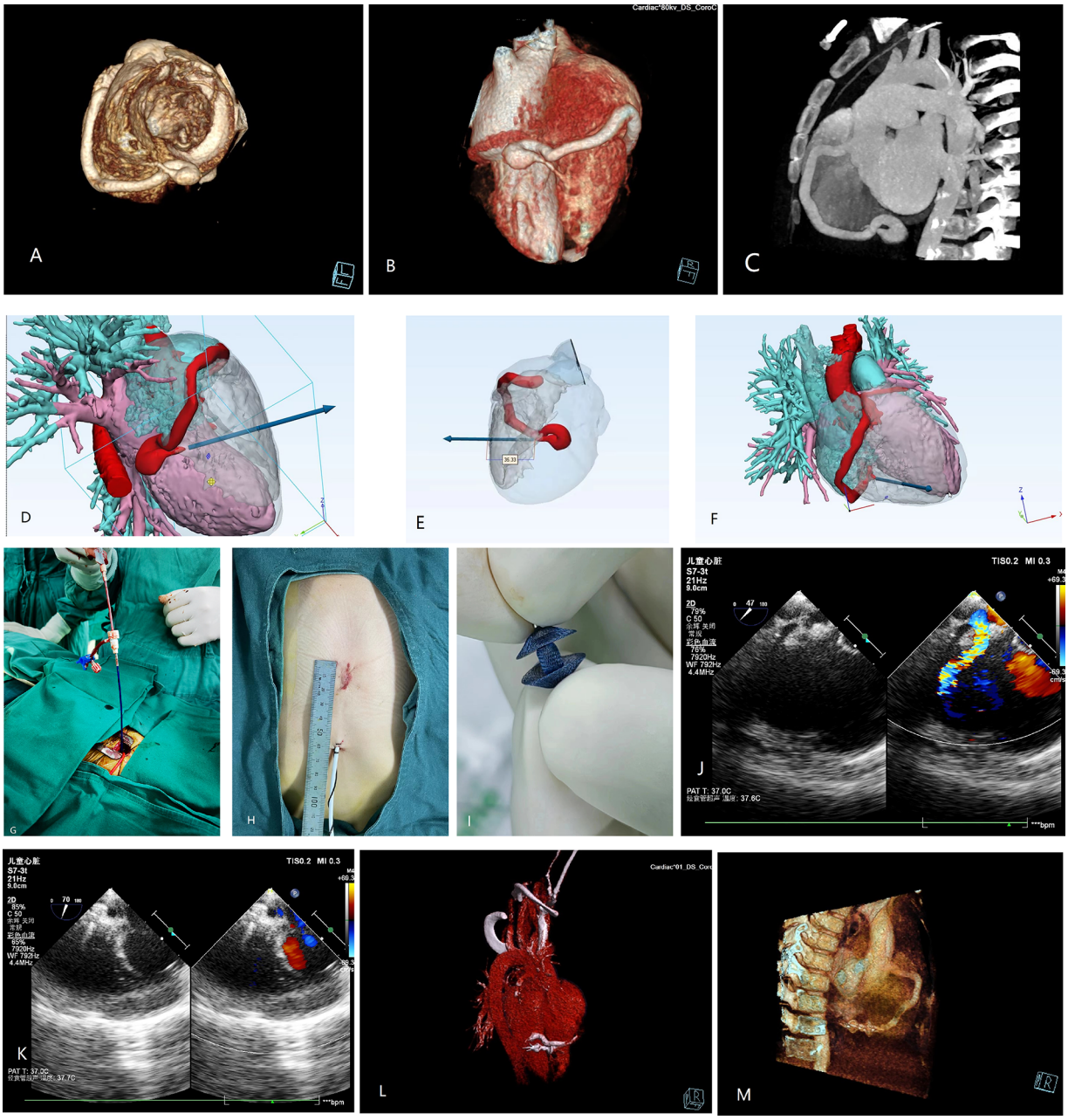

FIGURE 1
Preoperative 3D (A,B) and 2D (C) computed tomography (CT) images. (D–F) 3D images simulating the fistula drainage position and allowing reverse extrapolation of the puncture site and depth on the right ventricular free wall. (G) Surgical procedure. (H) Access incision. (I) Occluder. (J,K) Intraoperative ultrasound image for identifying the direction of blood flow and determining the direction of sheath guidance. Postoperative 3D (L) and 2D (M) CT images of the successfully occluded coronary artery fistula.

atrioventricular groove to the base of the heart, making a 360° rotation before draining into the RV, with a posterior descending branch evident at the lateral wall of the fistula. Preoperative simulation using computer-based 3D imaging (Figure 1D–F) was performed to determine the puncture site on the right ventricular free wall, as well as the sheath direction and insertion depth. The case was discussed during a multidisciplinary meeting and a decision for elective minimally invasive hybrid surgery was taken at the age of 3.8 years old (15 kg/100 cm).

2.1 Surgical procedure

After obtaining the parents' consent, the procedure was undertaken in the operating theater under transesophageal echocardiography (TEE) guidance. After systemic heparinization and antibiotic prophylaxis, a 2 cm incision was made at the inferior medial end of the sternum, with dissection of the inferior sternum layer by layer. The pericardium was incised and suspended to expose the anterior wall of the RV. The puncture site was identified using a combination of preoperative 3D imaging and intraoperative TEE (Figure 1D–F, Figure 1J–K). A purse-string suture was placed at the puncture site, followed by puncture and insertion of the guidewire (4-F) and sheath (1.17 mm). The TEE operator guided the sheath into the fistula, and a 4 × 4 mm Amplatzer Duct Occluder II (ADO II) (Abbott, USA) was inserted along the sheath until it was implanted. The occluder was released after a multi-view ultrasound confirmed no residual shunt or abnormalities (Figure 1G–K). The procedure lasted 30 min. The patient was extubated on the first postoperative day and discharged on the fourth postoperative day under oral heparin. Repeat examinations showed normal results (Figure 1L, M). Follow-up at 1 month, 2 months, 6 months, and 1 year revealed the occluder was well-positioned with no significant abnormalities observed.

3 Discussion

CAFs are a very rare congenital heart disease. Small CAFs may close spontaneously over time, whereas medium and large fistulas may expand and lead to complications such as coronary artery dilatation, aneurysm, atherosclerosis, myocardial infarction, and cardiac arrhythmia (2, 5, 6). Therefore, early treatment is recommended once diagnosed (7).

Traditional surgery and interventional closure are the main methods used to treat CAFs (8). However, traditional surgery involves performing a sternotomy, with some patients requiring extracorporeal circulation, which is highly traumatic and prone to risk of complications. Although interventional closure does not require a sternotomy or extracorporeal circulation (2), it has certain anatomical structure requirements (4, 9), and shortcomings such as radiation damage, vascular damage, and contrast allergy can occur intraoperatively. Furthermore, it carries a chance of postoperative complications, such as device displacement, myocardial infarction, and thrombosis (4). With recent advances in medical technology, minimally invasive hybrid closure has been increasingly employed in cardiac surgery, with studies showing it to be a safe and effective treatment approach for CAFs (10). Not only does the procedure avoid the need for extracorporeal circulation and x-ray irradiation, but it also has a wider scope of application and greatly reduces the occurrence of intraoperative and postoperative complications (3, 10, 11). However, minimally invasive hybrid closure procedure demands a high level of proficiency frolm the operator along with meticulous of the appropriate occluder. Physicians must carefully choose the size and model of the device based on the patient's unique condition, ensuring precise delivery to the defect site during the procedure. The timing and location of device release are critical for a successful outcome. While the procedure offers benefits such as reduced trauma and faster recovery, it is not without risks. potential complications may include device detachment, migration, thrombosis formation, and cardiac perforation. In our patient, the right coronary artery was dilated, extending from the atrioventricular groove to the base of the heart and turning a 360° rotation before draining into the right ventricle, with the posterior descending branch positioned at the lateral wall of the fistula. Because both the traditional surgical treatment and interventional closure options would have led to difficulties in this young patient, we decided that minimally invasive hybrid closure would be more suitable for him and performed the preoperative evaluation of cardiac anatomy using 3D imaging.

Three-dimensional imaging refers to the computer-aided design of 3D digital models that can simulate the visual experience of the human eye for real-world objects, allowing images or videos to be captured on visual media. Since we can only parse images on a two-dimensional level and lack 3D imagination, our ability to grasp anatomical details to achieve fine operation is limited. Therefore, we used 3D imaging to guide the surgery, which not only enabled preoperative simulation and surgical training but also helped the patient's family to better understand the treatment options and surgical methods during the communication process and gain an understanding of our clinical work. Additionally, 3D imaging allows us to tailor personalized anatomical structures for the patient, observe anatomical details more intuitively and accurately, and attain a good basis for surgical planning and surgical simulation (12, 13). This technology has also been applied more extensively for the guidance of cardiac surgery. Nazario (2022) reported the use of 3D printing in the surgical treatment of left ventricular aneurysms in 2022 (14). In our case, we combined this technique with preoperative cardiac CTA to construct the 3D anatomical structure of the heart as well as clarify the origin, course, and location of the CAF, construct a coordinate system on 3D imaging, reversely extrapolate the location of the puncture site, predict the sheath guidance direction and insertion depth, and formulate a surgical plan preoperatively. These actions enabled the safe and efficient implementation of the procedure.

Three-dimensional imaging-assisted minimally invasive hybrid closure can successfully be used to treat anatomically complex CAFs. To the best of our knowledge, this is the first report on the successful implementation of 3D imaging-assisted hybrid closure for a CAF in a pediatric patient. Using preoperative 3D

imaging, we were able to intuitively visualize the location of fistula drainage, infer the direction of blood flow, reverse extrapolate the puncture site on the right ventricular free wall, and predict the direction of sheath guidance, thereby avoiding blind penetration of the puncture site without first clarifying the direction of puncture during the surgical process. When combined with intraoperative TEE, we were able to rapidly identify the fistula and predict the depth of sheath insertion, thereby preventing complications arising from damage to the posterior ventricular wall and coronary artery due to excessively deep sheath insertion.

4 Conclusion

For CAFs with complex anatomical structures, such as tortuous courses, preoperative 3D imaging to visualize cardiac anatomy can significantly enhance the success of minimally invasive hybrid closure procedures and reduce the risk of complications in small children. This technique holds great potential for improving outcomes and should be further promoted in clinical practice.

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

The studies involving humans were approved by Dalian Municipal Women and Children's Medical Center (Group). The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation in this study was provided by the participants' legal guardians/next of kin. Written informed consent was obtained from the individual(s), and minor(s)' legal guardian/next of kin, for the publication of any potentially identifiable images or data included in this article.

Author contributions

YC: Writing – original draft, Writing – review & editing, Conceptualization, Investigation, Software, Validation, Visualization. JL: Writing – original draft, Writing – review & editing, Conceptualization, Investigation, Methodology, Project administration. XL: Writing – review & editing, Data curation, Formal Analysis, Project administration, Validation. PC: Writing – review & editing, Project administration, Resources, Supervision, Validation, Visualization. PW: Writing – review & editing, Data curation, Formal Analysis, Project administration, Resources, Software, Supervision, Visualization. LM: Writing – review & editing, Conceptualization, Data curation, Methodology, Project administration, Validation. YL: Writing – review & editing, Conceptualization, Data curation, Formal Analysis, Methodology, Project administration, Resources, Software, Supervision, Visualization.

Funding

The author(s) declare that no financial support was received for the research, authorship, and/or publication of this article.

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

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fcvm.2024.1439263/full#supplementary-material

References

1. Torres S, Vasconcelos M, Tavares Silva M, Moreira J, Silva JC, Macedo F. Coronary artery fistulas: a 12-year single-center experience. *Rev Port Cardiol.* (2022) 41(10):843–50. English, Portuguese. doi: 10.1016/j.repc.2021.06.024

2. Buccheri D, Chirco PR, Geraci S, Caramanno G, Cortese B. Coronary artery fistulae: anatomy, diagnosis and management strategies. *Heart Lung Circ.* (2018) 27 (8):940–51. doi: 10.1016/j.hlc.2017.07.014

3. Song L, Dong Y, Wang B, Li J, Qi X, Gao X, et al. The evolution of the minimally invasive approach and conventional median sternotomy for coronary artery Fistula correction. *Heart Lung Circ.* (2019) 28(8):1277–82. doi: 10.1016/j.hlc.2018.06.1052

4. Christmann M, Hoop R, Dave H, Quandt D, Knirsch W, Kretschmar O. Closure of coronary artery fistula in childhood: treatment techniques and long-term follow-up. *Clin Res Cardiol.* (2017) 106(3):211–8. doi: 10.1007/s00392-016-1041-6

5. Bolukcu A, Topcu AC, Sensoz Y, Kayacioglu I. Coronary artery fistulae involving both right and left coronary arteries. *Ann Thorac Surg*. (2017) 103(3):e261–2. doi: 10.1016/j.athoracsur.2016.08.050

6. Pu L, Li R, Yang Y, Liu G, Wang Y. Right coronary artery coronary sinus fistula with coronary sinus ostium stenosis. *Echocardiography*. (2017) 34(7):1102–4. doi: 10.1111/echo.13559

7. Karazisi C, Eriksson P, Dellborg M. Coronary artery fistulas: case series and literature review. *Cardiology*. (2017) 136(2):93–101. doi: 10.1159/000447445

8. Armsby LR, Keane JF, Sherwood MC, Forbess JM, Perry SB, Lock JE. Management of coronary artery fistulae. Patient selection and results of transcatheter closure. *J Am Coll Cardiol*. (2002) 39(6):1026–32. doi: 10.1016/S0735-1097(02)01742-4

9. Al-Hijji M, El Sabbagh A, El Hajj S, AlKhouli M, El Sabawi B, Cabalka A, et al. Coronary artery fistulas: indications, techniques, outcomes, and complications of transcatheter Fistula closure. *JACC Cardiovasc Interv*. (2021) 14(13):1393–406. doi: 10.1016/j.jcin.2021.02.044

10. Liu H, Mei J, He Y, Jiang Z, Tang M. Minimally invasive surgery for coronary-pulmonary artery Fistula via parasternal minithoracotomy. *Heart Lung Circ*. (2019) 28(4):e79–82. doi: 10.1016/j.hlc.2018.04.307

11. Song L, Wang B, Jin J, Hua Z, Yu H, Tao L. Minimally invasive approach for coronary artery fistula correction. *Interact Cardiovasc Thorac Surg*. (2017) 24(2):229–33. doi: 10.1093/icvts/ivw316

12. Salavitabar A, Figueroa CA, Lu JC, Owens ST, Axelrod DM, Zampi JD. Emerging 3D technologies and applications within congenital heart disease: teach, predict, plan and guide. *Future Cardiol*. (2020) 16(6):695–709. doi: 10.2217/fca-2020-0004

13. Garg R, Zahn EM. Utility of three-dimensional (3D) modeling for planning structural heart interventions (with an emphasis on valvular heart disease). *Curr Cardiol Rep*. (2020) 22(10):125. doi: 10.1007/s11886-020-01354-6

14. Carrabba N, Buonamici F, Furferi R, Carfagni M, Vannini M, Valenti R, et al. Case report: three-dimensional printing model for surgical planning of left ventricular aneurysm: evolution toward tailoring surgery. *Front Cardiovasc Med*. (2022) 9:852682. doi: 10.3389/fcvm.2022.852682

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TYPE Case Report
PUBLISHED 04 December 2024
DOI 10.3389/fcvm.2024.1507222

OPEN ACCESS

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RECEIVED 07 October 2024
ACCEPTED 14 November 2024
PUBLISHED 04 December 2024

CITATION
Aigumov RN, Donakanyan SA, Merzlyakov VY, Skopin AI, Baichurin RK, Panagov ZG, Sizhazhev EM, Shvartz VA and Golukhova EZ (2024) Geometric reconstruction of the left ventricle on a beating heart through a minimally invasive approach from the left anterolateral thoracotomy: case report. Front. Cardiovasc. Med. 11:1507222. doi: 10.3389/fcvm.2024.1507222

Geometric reconstruction of the left ventricle on a beating heart through a minimally invasive approach from the left anterolateral thoracotomy: case report

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Despite the widespread use of mini-invasive treatment methods in cardiac surgery, their use in post-infarction myocardial aneurysms of the left ventricle is not of frequent occurrence. In this clinical case, we used left anterolateral thoracotomy and "eating heart" technique to correct the post-infarction left ventricle aneurysm with ventricular thrombosis using the Dor method in a 66-year-old patient. This technique created opportunity to perform safely and effective the planned reconstruction of the left ventricle with less trauma, as well as to ease the postoperative course and recovery of the patient, reduce hospitalization time.

KEYWORDS

left ventricle reconstruction, mini-invasive approach, cardiovascular pathology, ventricular aneurysm, cardiovascular research, case report

Introduction

Size enlargement, shape distortion, and dysfunction of the left ventricle (LV) due to necrosis and scarring are characteristic signs of postinfarction left ventricular aneurysm (PLVA). These pathological processes are the main target of surgical intervention, the purpose of which is to isolate devitalized tissues with restoration of the elliptical shape and proper dimensions of the LV (1). The Dor procedure is among the top recognized surgical techniques used for this purpose.

The standard Dor procedure is a geometric reconstruction of the LV performed through a median sternotomy under conditions of cardioplegic arrest (2).

The advancement of this technique has led to its implementation on a beating heart under cardiopulmonary bypass (CPB) (3–6).

The world literature includes just a few publications dedicated to the experience of performing LV reconstruction through left anterior minithoracotomy in a stopped heart (7–9).

In this report, we describe a case of a successful Dor procedure performed under conditions of minimal access and parallel perfusion.

Case description

A male patient (66 years of age, weighing 80 kg) was sent to our Center with signs of the functional class III heart failure and diagnosed with thrombosed PLVA. Upon admission to the department, the diagnosis was confirmed by electrocardiography, echocardiography (EchoCG), and ventriculography. The ECG is specific, it is shown in Figure 1. EchoCG data revealed an enlarged LV with the left ventricular end-diastolic volume (LV EDV) of 177 ml, end-sistolic volume (LV ESV)—116.8 ml, EDV index—88.5 ml/m^2, ESV index—58.4 ml/m^2, a change in its shape (left ventricular sphericity index of 0.65 in diastole and apical conicity index of 0.93 in systole), areas of hypo- and akinesia with the formation of a thrombosed aneurysm of anterolateral segment with transition to the apex (aneurysm volume of 56 ml, thrombus size of 17 × 29 mm), and also a reduction in global contractility of the LV myocardium characterized by the left ventricular ejection fraction (LVEF) of 34%. Left ventriculography in two projections confirmed the fact of pronounced negative remodeling of the LV in the form of an aneurysm with thrombus formation (Figures 2A,B).

According to the results of coronary catheterization, occlusion of the left anterior descending branch of the left coronary artery (LAD LCA) was noted without proper filling of the lumen along intersystem and intrasystem collaterals (Figure 2C).

It should be noted that this patient had a history of cerebral circulatory disorders (transient ischemic attack).

Considering the high functional class of chronic heart failure, the large volume of the aneurysm, LV dysfunction with LVEF under 35%, as well as the presence of a life-threatening thrombus in the left ventricular lumen, the only predictably effective treatment method was open-heart bypass surgery.

Surgical intervention

The heart was accessed through a left anterolateral minithoracotomy in the fifth intercostal space (Figure 3A). The surgery was performed under conditions of parallel perfusion with the connection of peripheral CPB through the left femoral vein and femoral artery. The venous cannula was installed in the right atrium closer to the superior vena cava under the control of transesophageal EchoCG. After opening the pericardium and its fixation, we performed a ventriculotomy, removed the thrombus (Figure 3B), and then, under full visualization of the boundaries of the viable myocardium (Figure 3C), we carried out a geometric reconstruction of the LV following the Dor procedure, using a synthetic patch made of Dacron (polyethylene terephthalate) (Figure 3D).

The duration of CPB was 36 min. The time of artificial lung ventilation was 11 h. There was no need for cardiotonic agents. The next day after the surgery, the patient was transferred to the general ward from the intensive care unit, and verticalization of the patient was attempted. He was discharged in stable condition on the fifth day. Basic EchoCG data at discharge were as follows: left ventricular end-systolic volume, LV EDV = 135.3 ml (EDV index—67.6 ml/m^2), LV ESV = 62.0 ml (ESV index—31 ml/m^2); LVEF = 54.2%. TEE after the surgery shows physiologically more correctly formed LV geometry (Figure 4).

Discussion

This clinical case reports the possibility of adequately performing complex surgical reconstruction on the LV by means of a minithoracotomy and on a beating heart under CPB conditions.

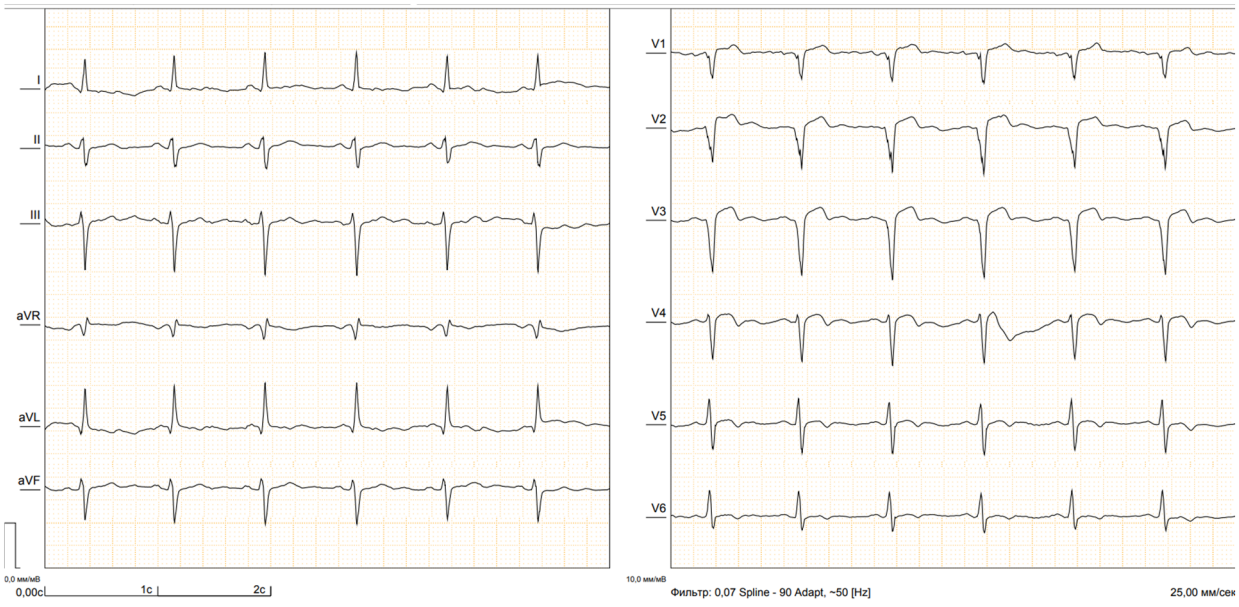

FIGURE 1
Electrocardiogram of the patient.

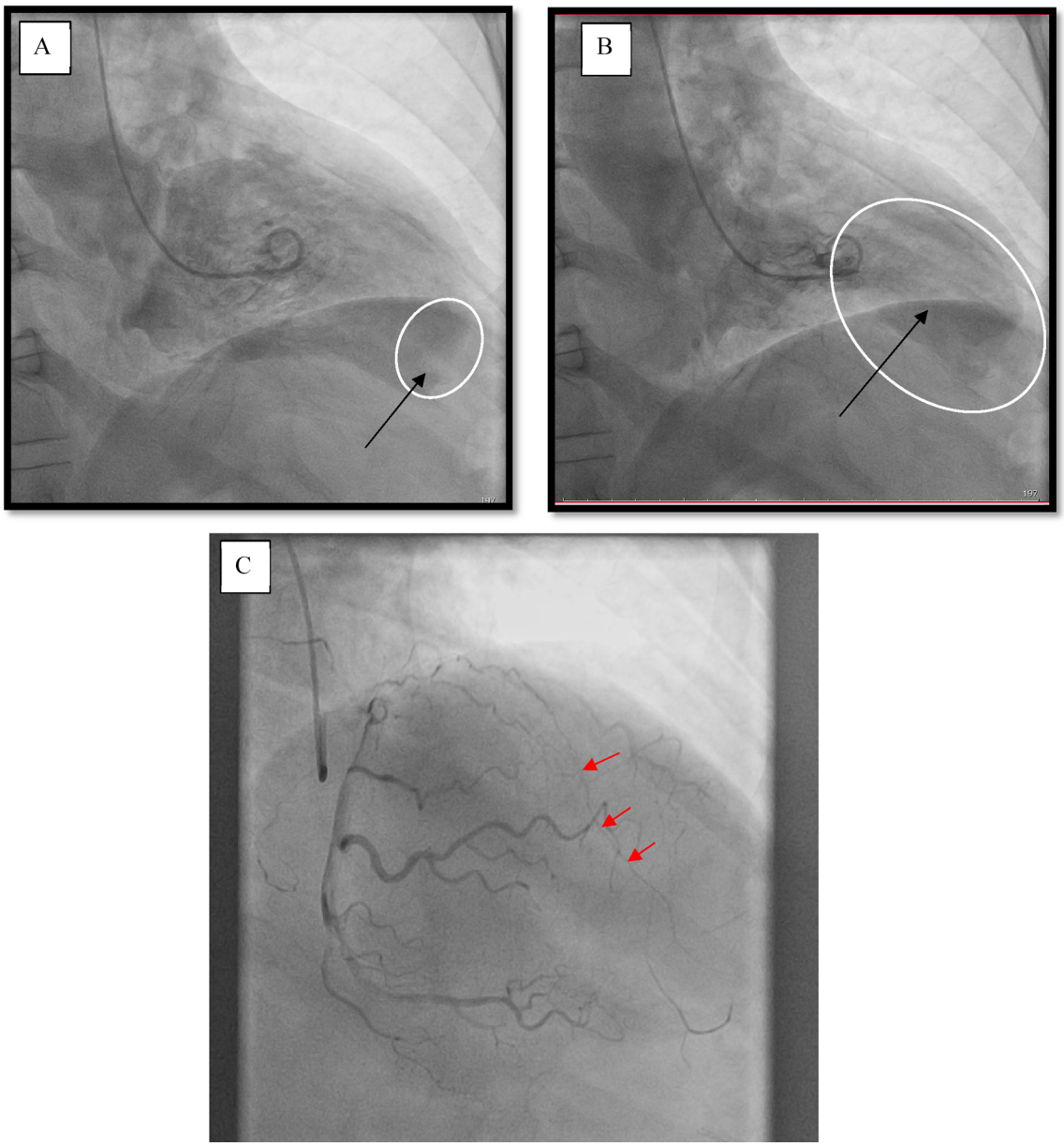

FIGURE 2
Left ventriculography. **(A)** Diastole, the arrow points at a thrombus at the left ventricle apex. **(B)** Systole, the arrow indicates the area of the aneurysm. **(C)** Coronary catheterization. The arrows point at the occlusion of the left anterior descending branch of the left coronary artery (LAD LCA).

The Dor procedure was first developed in the mid-1980s as a novel method for surgical treatment of PLVA. It involves endoventricular LV repair using a circumferential pursestring suture and a circular patch (2). We performed this complex intervention under standard conditions: median sternotomy and cardioplegic arrest of the heart (2). To date, this approach remains the gold standard worldwide in the surgery of LV aneurysms. However, over the past two decades, interest in minimally invasive procedures in cardiac surgery has increased, which has led to an expansion of the arsenal of surgical approaches, thereby eliminating traditional, more traumatic surgical conditions.

Considering possible complications associated with cardioplegic arrest of the heart, the method of surgical treatment of PLVA on a beating heart has become an effective alternative. Besides, it provides better visualization of the boundaries of the viable myocardium (3–6).

As part of achieving minimal invasiveness, the possibilities of using various surgical approaches instead of the more traumatic median sternotomy have expanded. In this case, the optimal mini-access is a left anterolateral thoracotomy.

The techniques of MIDCAB as a form of off-pump coronary artery bypass surgery and MICS CABG as an advanced minimally invasive technique for multi-vessel bypass, performed

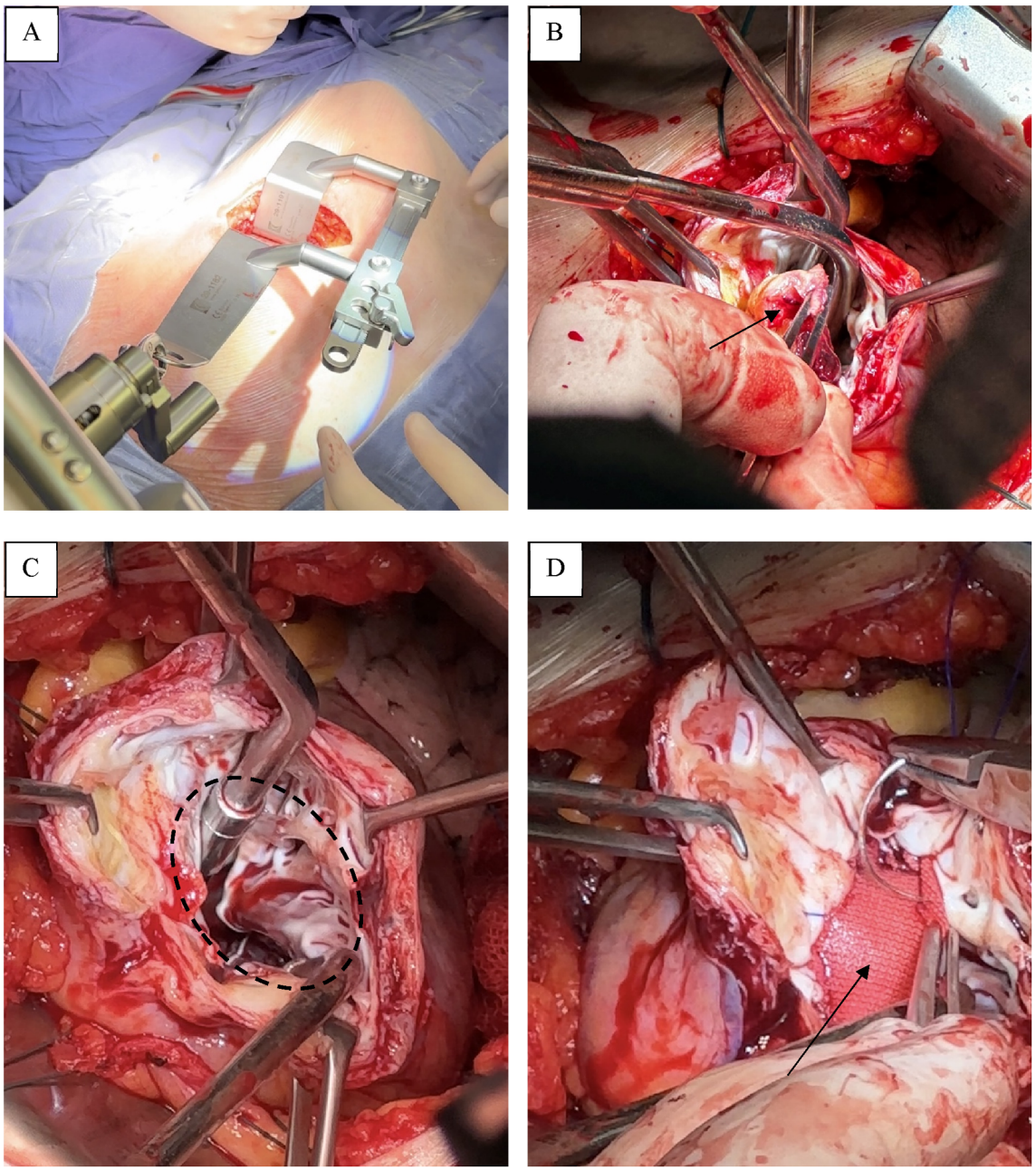

FIGURE 3
Operative stage. **(A)** A left anterolateral minithoracotomy in the fifth intercostal space. **(B)** Thrombus. **(C)** Boundaries of the viable myocardium. **(D)** Synthetic patch made of Dacron.

through this approach, demonstrate the option of performing technical actions with good results not only in the area of the apex and anterior wall of the heart, but virtually over its entire surface, which makes left anterior minithoracotomy highly attractive (10).

Previously, the possibility, effectiveness and safety of the LV surgical reconstruction through left anterior minithoracotomy was verified by colleagues from Europe, but all procedures were performed under conditions of cardioplegia and aortic cross-clamping through the port-access approach (7, 8) or additional thoracotomy (9).

Hence, our surgical strategy was to combine these two attractive minimally invasive approaches to the treatment of LV aneurysms.

LV thrombosis was also an important factor determining the indications for this operation, in addition to the high functional class of chronic heart failure and a large LV aneurysm, which occupied almost a third of the total LV volume with decreased global myocardial contractility. This factor was regarded as a life—threatening source of thromboembolic complications, taking into account patient's aggravating neurological history—a postponed episode of a cerebrovascular event.

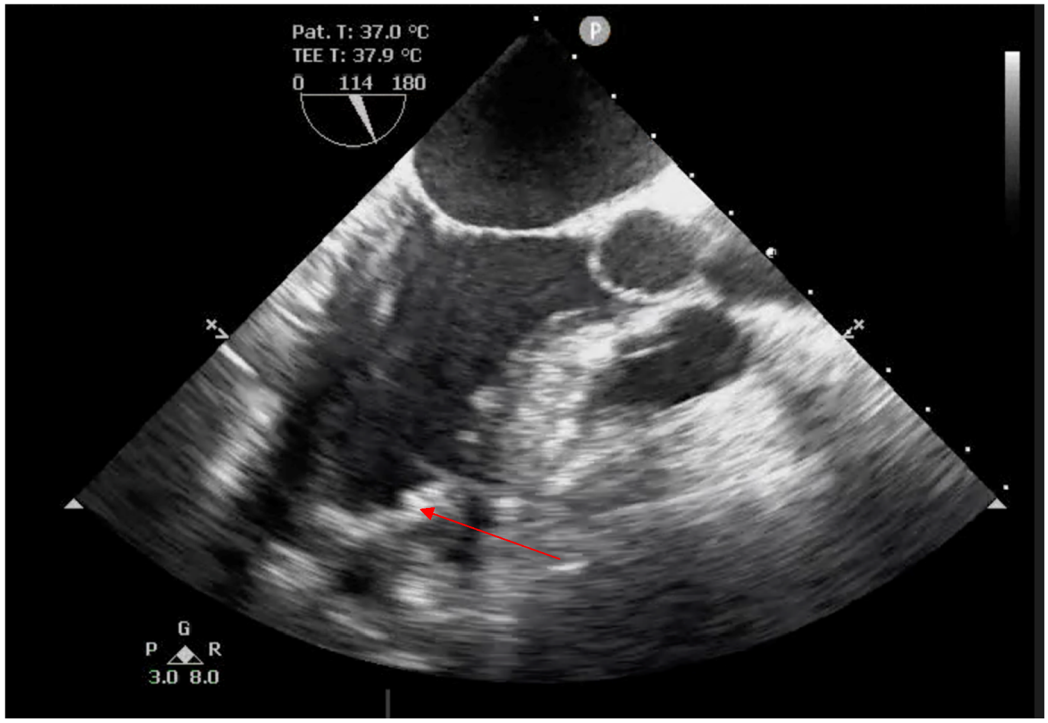

FIGURE 4
TEE after the surgery. LV, systole. The arrow points at the patch.

During the preoperative planning of the intervention strategy, taking into account the need for only isolated treatment of LV aneurysm without CABG and the patient's preference, we chose the method of surgical intervention through a minimally invasive approach on a beating heart, which was a pioneering method in Russia. Furthermore, we did not find any publications in the worldwide reference databases dedicated to the experience of performing such operations.

The condition in which the venous cannula reaches the lumen of the superior vena cava, as well as vacuum-assisted venous drainage, are very important, because this allows for maximum LV unloading and, consequently, provides optimal and safe visualization.

Performing surgery on a beating heart made it possible to more clearly visualize the demarcation zone and properly isolate the scarred part of the aneurysm.

It is necessary to keep the patient's head slightly lowered and keep sufficient perfusion pressure to prevent the air embolism during the main stage of the operation. After careful removal of the thrombus, a thorough examination of the LV cavity for the presence of foreign bodies was performed. Further, enough blood is released from this cavity under pressure to completely evacuate possible hidden emboli. The presence of aortic insufficiency is a contraindication for performing this procedure. The final sutures on the patch are applied in conditions of a filled heart after complete deaeration through the left hole, then sealing with auto-tissues is achieved.

The technique of a "Beating heart" allows minimizing the risks of developing of the small cardiac output syndrome associated with an excessive decrease of the LV cavity volume, since the boundary between a viable and non-viable myocardium is clearly visualized intraoperatively. This boundary is a guideline for preoperative intervention design based on echocardiography, ventriculography/MRI data. Such a concept makes it possible to achieve not only the optimal volume, but also the shape of the LV (11).

The outcome of this surgery is impressive. This method provides an early mobilization of the patient (already on the first day after surgery) and his or her early discharge (on the fourth or fifth day). Values of indicators obtained with a control EchoCG imply proper isolation of the affected area, while maintaining optimal LV volume. There were no signs of heart failure in the postoperative period and, accordingly, there was no need for infusion of cardiotonic preparations, which is one of the advantages of the intervention without cardioplegia.

The skills of minimally invasive CABG, as well as the accumulation of experience in minimally invasive LV reconstruction on a beating heart, will expand the potential of combined intervention for complex coronary artery disease in the future.

Further studies using larger sample sizes and long-term outcome studies are needed.

Conclusion

Left anterolateral minithoracotomy is an approach that allows proper surgical reconstruction of the LV on a beating heart with good results. This approach provides optimal surgical field

visibility and can be used in patients with PLVAs. Future large-scale studies analyzing long-term results will allow researchers to draw unambiguous conclusions.

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 humans were approved by Local Ethics Committee of Bakulev National Medical Research Center for Cardiovascular Surgery of the Russian Ministry of Health. The studies were conducted in accordance with the local legislation and institutional requirements. The 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

RA: Conceptualization, Data curation, Investigation, Methodology, Resources, Software, Supervision, Visualization, Writing – original draft. SD: Data curation, Methodology, Resources, Supervision, Visualization, Project administration, Validation, Writing – review & editing. VM: Data curation, Validation, Visualization, Writing – review & editing. AS: Data curation, Visualization, Formal Analysis, Investigation, Software, Supervision, Writing – original draft. RB: Data curation, Software, Visualization, Writing – original draft. ZP: Data curation, Software, Visualization, Writing – original draft, Supervision. ES: Data curation, Software, Visualization, Writing – original draft. VS: Project administration, Writing – review & editing. EG: Funding acquisition, Project administration, Resources, Supervision, Writing – review & editing.

Funding

The author(s) declare that no financial support was received for the research, authorship, and/or publication of this article.

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.

Generative AI statement

The authors declare that no Generative AI was used in the creation of this manuscript.

Publisher's note

References

1. Ruzza A, Czer LSC, Arabia F, Vespignani R, Esmailian F, Cheng W, et al. Left ventricular reconstruction for postinfarction left ventricular aneurysm: review of surgical techniques. *Tex Heart Inst J.* (2017) 44(5):326–35. doi: 10.14503/THIJ-16-6068

2. Dor V, Saab M, Coste P, Kornaszewska M, Montiglio F. Left ventricular aneurysm: a new surgical approach. *Thorac Cardiovasc Surg.* (1989) 37(1):11–9. doi: 10.1055/s-2007-1013899

3. Yu Y, Gu CX, Wei H, Liu R, Chen CC, Fang Y. Repair of left ventricular aneurysm during off-pump coronary artery bypass surgery. *Chin Med J (Engl).* (2005) 118 (13):1072–5.

4. Wang X, He X, Mei Y, Ji Q, Feng J, Cai J, et al. Early results after surgical treatment of left ventricular aneurysm. *J. Cardiothorac. Surg.* (2012) 7:126. doi: 10.1186/1749-8090-7-126

5. Bokeria LA, Merzlyakov VY, Skopin AI, Klyuchnikov IV, Mammadova SK. Immediate results of left ventricular reconstruction on a contracting heart. *Russ J Thorac Cardiovasc Surg.* (2011) 4:36–8.

6. Tarasov DG, Chernov II, Molochkov AV, Pavlov AV. Experience of post-infarction left ventricular aneurysm reconstruction with on-pump beating heart technique. *Kazan Med J.* (2019) 100(3):500–4 (in Russ.). doi: 10.17816/KMJ2019-500

7. Alloni A, Rinaldi M, Gazzoli F, D'Armini AM, Viganò M. Left ventricular aneurysm resection with port-access surgery: a new mini-invasive surgical approach. *Ann Thorac Surg.* (2003) 75(3):786–9. doi: 10.1016/S0003-4975(02)04560-5

8. Totaro P, Alloni A, Cattadori B, Monterosso C, Degani A, Maurelli M, et al. Minimally invasive heart-port assisted technique as standard first choice approach for left ventricle endoplasty: a ten-year single-center experience. *Ann Thorac Surg.* (2010) 89(4):1200–3. doi: 10.1016/j.athoracsur.2010.01.035

9. Babliak O, Demianenko V, Marchenko A, Babliak D, Melnyk Y, Stohov O, et al. Left anterior minithoracotomy as a first-choice approach for isolated coronary artery bypass grafting and selective combined procedures. *Eur J Cardiothorac Surg.* (2023) 64 (2):ezad182. doi: 10.1093/ejcts/ezad182

10. McGinn JT Jr, Usman S, Lapierre H, Pothula VR, Mesana TG, Ruel M. Minimally invasive coronary artery bypass grafting: dual-center experience in 450 consecutive patients. *Circulation.* (2009) 120(11 Suppl):S78–84. doi: 10.1161/CIRCULATIONAHA.108.840041

11. Bockeria LA, Sokol'skaya NO, Kopylova NS, Alshibaya MD. Method of left ventricle end-diastolic volume determination during geometric left ventricle reconstruction in patients with postinfarction aneurysm. The bulletin of Bakoulev center. *Cardiovasc Dis.* (2019) 20(11–12): 1042–53 (in Russ.). doi: 10.24022/1810-0694-2019-20-11-12-1042-1053

frontiers | Frontiers in Cardiovascular Medicine

TYPE Case Report
PUBLISHED 11 December 2024
DOI 10.3389/fcvm.2024.1471686

OPEN ACCESS

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RECEIVED 28 July 2024
ACCEPTED 25 November 2024
PUBLISHED 11 December 2024

CITATION
Yang Y, Liu X, Liu J, Shi J and Li H (2024) Aortic valve replacement in a bicuspid aortic valve patient followed by reoperation for ascending aorta rupture: a case report.
Front. Cardiovasc. Med. 11:1471686.
doi: 10.3389/fcvm.2024.1471686

Aortic valve replacement in a bicuspid aortic valve patient followed by reoperation for ascending aorta rupture: a case report

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Bicuspid aortic valve (BAV), the most common congenital cardiac anomaly, predisposes individuals to aortic stenosis and regurgitation due to valve degeneration. Abnormal hemodynamics, arterial wall characteristics, and genetic factors contribute to ascending aorta dilatation, potentially leading to severe complications like aortic dissection. Presently, the most recent guidelines propose that individuals with BAV requiring valve replacement due to valve dysfunction should undergo simultaneous replacement of the ascending aorta when the diameter of aortic dilatation exceeds 4.5 cm. A 60-year-old female patient previously underwent mechanical aortic valve replacement and ascending aortoplasty at our center due to aortic stenosis and a 4.3 cm diameter ascending aorta. In the sixth postoperative year, she was readmitted due to ascending aorta rupture, resulting in blood entering the right atrium and causing acute right heart failure. We performed repair of the rupture and concomitant ascending aorta replacement to prevent further exacerbation of the patient's condition.

KEYWORDS

ascending aortic dilatation, aortic rupture, cardiac reoperation, bicuspid aortic valve, aortic valve replacement

Introduction

The bicuspid aortic valve (BAV) represents a prevalent congenital malformation arising from the fusion of the two aortic valve cusps in the trileaflet aortic valve (TAV). Apart from the symptoms attributable to the valve, such as stenosis and valvular insufficiency, the presence of abnormal hemodynamics amplifies the risk of aortic dilatation by 86% (1). According to research statistics, although the overall incidence of aortic dissection in patients with BAV is relatively low, it remains significantly higher than in the general population (2). Consequently, in individuals who present with BAV along with ascending aortic dilatation, determining the optimal timing and surgical approach is highly important. Previous research findings indicate that following aortic valve replacement in BAV patients, the probability of aortic expansion diminishes, approaching that observed in TAV patients. Simultaneously, the prevailing perspective regarding aortic replacement in BAV patients currently tends toward a conservative

approach. Aortic replacement is deemed reasonable only in situations where the BAV is coupled with valvular dysfunction requiring valve replacement and the diameter of aortic dilatation exceeds 4.5 cm (3). We report a rare case of rupture of the ascending aorta into the right atrium at 6 years after aortic valve replacement and ascending aortoplasty.

Case description

A 66-year-old female patient was identified six years ago with adhesions at the junction of the left and right coronary valves of the trileaflet aortic valve, resulting in a bicuspid deformity and severe stenosis of the aortic valve. Simultaneously, there was concurrent aneurysmal dilatation of the ascending aorta. Upon admission, the patient did not manifest any discernible symptoms. During physical examination, her heart rate was recorded at 84 beats per/minute, and her blood pressure was measured at 143/101 mmHg. Transthoracic echocardiography revealed an aortic valve opening area of approximately 0.9 cm^2, a peak systolic flow rate of 4.5 m/s, a differential pressure gradient of 82 mmHg, a left ventricular ejection fraction of 63%, a proximal aortic diameter of approximately 4.3 cm, and normal morphology and activity of the mitral and tricuspid valves. Aortic computed tomography angiography (CTA) results show thickening of the aortic valve with minimal calcification and dilation of the ascending aorta, with a diameter of 4.4 cm. Additionally, mild stenosis was observed in the anterior descending branch of the right coronary artery at the opening on coronary arteriography. Following a clear surgical indication, we considered performing mechanical aortic valve replacement alongside ascending aortic replacement or ascending aortoplasty for the patient. Ultimately, the patient chose ascending aortoplasty due to personal factors. The surgery was performed through a median sternotomy under moderate hypothermic cardiopulmonary bypass. After cardiac arrest, the aortic valve was excised through the aortic incision, and a mechanical valve prosthesis was placed, confirming that the valve leaflets opened and closed properly. A tapered resection of the dilated aortic wall was performed along the aortic incision, and the aortic incision was closed with a continuous suture using 5–0 Prolene thread. Postoperative examination revealed optimal functioning of the mechanical valve. Moreover, the ascending aorta exhibited satisfactory flow. The patient was discharged 16 days postsurgery. During the six-year follow-up period, the patient attended regular outpatient reviews at our center, where laboratory tests and transthoracic echocardiography results showed no significant abnormalities. Coagulation function was maintained within the acceptable range required after mechanical valve replacement.

Six years after the operation, the patient sought admission to the emergency department of our hospital and presented with intermittent chest tightness and pain, coupled with weakness and dizziness. A transthoracic echocardiogram revealed a rupture on the right posterior wall of the ascending aorta extending into the right atrium. The ascending aorta exhibited aneurysmal dilation, with a proximal diameter of approximately 5.1 cm, indicating slight expansion compared to six years ago. The mechanical aortic valve exhibited normal function, characterized by systolic blood flow without significant acceleration, a peak flow rate of 3.3 m/s, and a differential pressure gradient of 44 mmHg. The left ventricular ejection fraction was measured at 65%. Additionally, the patient presented with a combination of moderately severe tricuspid insufficiency, moderate pulmonary hypertension, and a moderate to large amount of systolic regurgitant signal at the tricuspid orifice, featuring a peak flow rate of 3.1 m/s and a pressure gradient of 39 mmHg. Upon admission to the hospital, the patient underwent emergency room CTA of the thoracic and abdominal aorta. Imaging revealed limited bulging and protruding shadows measuring approximately 38*18 mm at the right edge of the lower section of the ascending aorta. The tip of this bulging structure was suspected to be connected to the right atrium, which appears to be flattened due to compression (Figures 1a,b), and a three-dimensional CTA likewise showed a significant rupture at the ascending aorta (Figure 1c). Moreover, the patient exhibited an international normalized ratio of 2.54 s, a prothrombin time of 24.9 s, and an elevated N-terminal pro-B-type natriuretic peptide level of 4030.0 pg/ml. Ultimately, owing to acute right heart failure resulting from the influx of blood from the ascending aorta into the right atrium, the patient was subsequently transferred to our cardiovascular surgery department for further treatment.

Following the confirmation of the absence of contraindications to surgery, we proceeded with ascending aortic replacement using a median sternotomy combined with a right inguinal incision. Subsequently, we initiated extracorporeal circulation by cannulation through the femoral artery, superior vena cava, and inferior vena cava. When circulatory arrest was achieved circulatory arrest through whole-body mild hypothermia, the surgical procedure commenced with removal of the pseudoaneurysm. Subsequently, the right atrial rupture was meticulously closed. The proximal portion of the ascending aorta was then trimmed, and the concluding phase involved the execution of ascending aortic replacement. Next, evaluation of tricuspid valve function was carried out through a right atrial incision, and the water injection test demonstrated the absence of discernible regurgitation. Following the closure of the right atrial incision, we disengaged from the extracorporeal circulation and meticulously closed the thoracic cavity using layer-by-layer sutures. Intraoperative transesophageal ultrasonography revealed unimpeded blood flow in the artificial vessel after ascending aortic replacement, patent coronary artery openings on both the right and left sides, and no discernible abnormalities subsequent to the repair of the right posterior wall breach of the ascending aorta. No discernible signal was detected during the diastolic phase of the aortic valve. Following the procedure, the patient was transferred to the cardiac care unit.

On the second postoperative day, a bedside transthoracic echocardiogram revealed the patent status of the artificial ascending aortic vessel, with a peak systolic flow at the aortic valve orifice measuring 2.6 m/s and a differential pressure gradient of 27 mmHg. Moreover, there was a normalization of

tricuspid valve morphology and activity. After the surgery, sections of the patients' aorta were subjected to pathological staining for analysis. Hematoxylin-eosin staining (Figure 2a), Masson's trichrome staining (Figure 2b), and Verhoeff-van Gieson elastic staining (Figure 2c) collectively revealed histological features indicative of smooth muscle cell disorganization, accompanied by a discernible decrease in elastin and collagen fiber density. Prior to discharge, the patient demonstrated satisfactory cardiac function without any cardiovascular complications. Subsequent regular reviews at our outpatient clinic revealed no significant abnormalities in the patient's condition.

Discussion

BAV is a highly heritable autosomal dominant genetic disorder, with disease progression, patients with BAV may experience aortic stenosis, aortic complications, infective endocarditis, and congestive heart failure (4). Long-term follow-up studies indicate that the probability of aortic dilation in BAV patients can be as high as nearly 50% (5). Furthermore, the risk of aortic aneurysm in BAV patients is significantly higher than that in the general population (2). Studies confirm that even without accompanying aortic valve stenosis or regurgitation, ascending aortic dilatation can be an early complication in BAV patients, which explains why ascending aortic dilatation can occur as early as the fetal period in these patients (6). A considerable percentage of individuals necessitating aortic valve replacement concurrently exhibit dilatation of the ascending aorta. If left unaddressed, this condition may give rise to potentially fatal aortic adverse events, as indicated by pertinent research findings (7). Winkler et al. paired 120 patients who underwent simple aortic valve replacement with 40 patients who underwent aortic valve

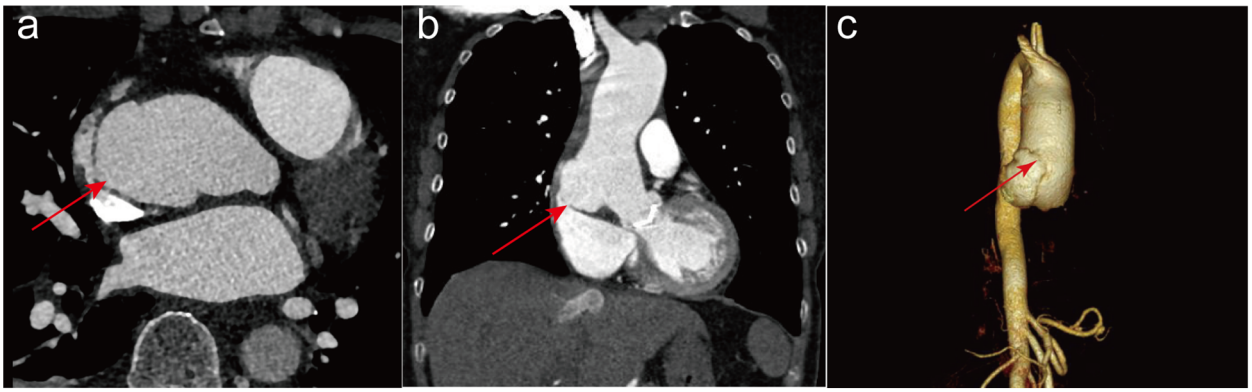

FIGURE 1
The imaging tests conducted on patients after admission to the hospital. Preoperative computed tomography angiography of the thoracic and abdominal aorta in both transverse (a) and longitudinal (b) sections showed a significant rupture at the ascending aorta, with the rupture connecting to the right atrium as indicated by the red arrow. Three-dimensional CTA with red arrows also shows a significant rupture in the ascending aorta (c).

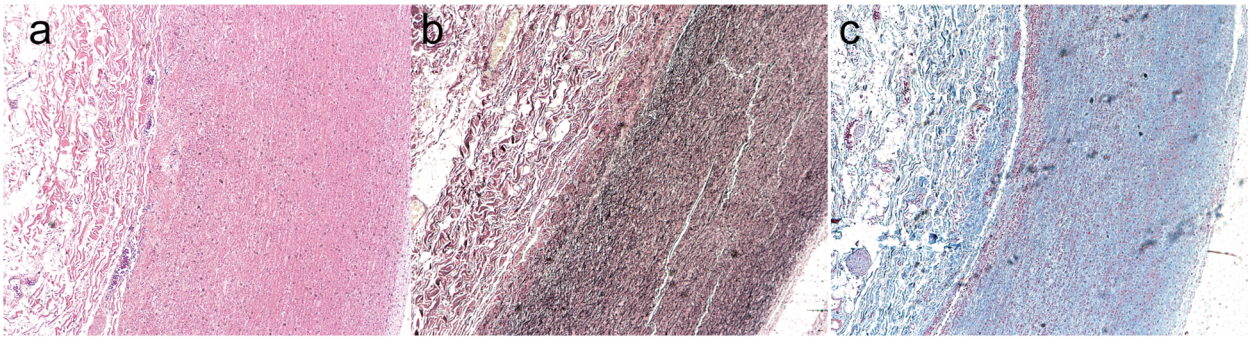

FIGURE 2
The postoperative patients' aorta underwent pathological section staining. Hematoxylin-eosin staining (a), Masson's trichrome staining (b), and Verhoeff-van Gieson elastic staining (c) collectively revealed histological features indicative of smooth muscle cell disorganization, accompanied by a discernible decrease in elastin and collagen fiber density.

replacement along with ascending aortic replacement, employing a propensity score matching ratio of 3:1. Their conclusive findings assert that the amalgamation of ascending aortic replacement does not augment surgical morbidity or mortality rates (8). In a parallel fashion, Idrees et al. employed propensity scores to pair 647 sets of patients, revealing that the execution of prophylactic aortic replacement concurrently with cardiac surgery involving mild concomitant aortic disease does not increase the risk of mortality (9). This outcome substantiates the safety associated with simultaneous aortic replacement during heart valve surgery. Furthermore, aortic lesions in patients with BAV exhibit abnormal hemodynamics and are predisposed to evolve into dilated aortic lesions in a more pronounced manner than those in TAV patients (10, 11). According to the 2022 American College of Cardiology/American Heart Association guidelines, surgical replacement of the ascending aorta is recommended for individuals with BAV and an aortic diameter equal to or exceeding 5.5 cm. Simultaneously, the guidelines posit that concurrent replacement of the ascending aorta is deemed a reasonable course of action for patients, necessitating valve replacement due to valve dysfunction, provided that the aortic diameter reaches or exceeds 4.5 cm (12). Nonetheless, empirical evidence has demonstrated that a substantial number of acute type A aortic dissections manifest at dimensions below the aforementioned threshold. Ascending aortic replacement may be considered in patients with BAV combined with ascending aortic dilatation, even if the aortic diameter is less than 5.0 cm (13). Moreover, magnetic resonance imaging studies have conclusively revealed a significantly greater prevalence of both ascending aortic diameter and ascending aortic dilatation in patients diagnosed with BAV than in those diagnosed with TAV (14). Investigations have indicated that individuals with BAV exhibit thinner elastic lamellae in the aorta along with a greater interlamellar distance when juxtaposed with patients harboring TAV (15). These structural distinctions may collectively contribute to accelerated progression of aortic pathology and increased vulnerability to adverse aortic events in individuals with BAV. The likelihood of adverse aortic events following isolated aortic valve replacement in individuals with mild to moderate ascending aortic dilatation is relatively low (16). However, due to the potential for aortic dissection following aortic dilation, prophylactic surgery remains necessary (17). In summary, even if the ascending aortic diameter in BAV patients has not reached the surgical threshold, surgeons may still reasonably consider ascending aortic replacement based on the individual patient's condition and actual clinical circumstances.

In accordance with the surgeon's clinical expertise and adherence to prevailing guideline criteria, none of the established surgical thresholds for ascending aortic replacement were met. Consequently, the chosen intervention involved aortic valve replacement concomitant with ascending aortoplasty. While ascending aortoplasty yields positive early-stage results, effectively slowing aortic diameter growth, and has the advantages of being safe and effective, with fewer complications and a lower mortality rate, patients who undergo BAV still have problems with re-expansion. Hence, a prolonged postoperative follow-up is necessary (18, 19).

Individuals with BAV and concurrent ascending aortic dilatation should undergo early surgical intervention. The selection of appropriate surgical modalities and personalized treatment plans is crucial for mitigating the risk of severe complications. This case also reminds us that more aggressive treatment of the dilated ascending aorta in practice may be a more sustainable and wise choice.

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 humans were approved by the Ethics Committee of Union Hospital, Tongji Medical College, Huazhong University of Science and Technology. The studies were conducted in accordance with the local legislation and institutional requirements. The human samples used in this study were acquired from primarily isolated as part of your previous study for which ethical approval was obtained. Written informed consent for participation was not required from the participants or the participants' legal guardians/next of kin in accordance with the national legislation and institutional requirements. 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

YY: Conceptualization, Data curation, Visualization, Writing – original draft. XL: Conceptualization, Methodology, Writing – review & editing. JL: Conceptualization, Methodology, Writing – review & editing. JS: Conceptualization, Project administration, Supervision, Writing – review & editing. HL: Conceptualization, Project administration, Supervision, Writing – review & editing.

Funding

The author(s) declare that no financial support was received for the research, authorship, and/or publication of this article.

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.2024.1471686/full#supplementary-material

References

1. Broberg CS, Therrien J. Understanding and treating aortopathy in bicuspid aortic valve. *Trends Cardiovasc Med*. (2015) 25:445–51. doi: 10.1016/j.tcm.2014.12.006

2. Michelena HI, Khanna AD, Mahoney D, Margaryan E, Topilsky Y, Suri RM, et al. Incidence of aortic complications in patients with bicuspid aortic valves. *JAMA*. (2011) 306:1104–12. doi: 10.1001/jama.2011.1286

3. Borger MA, Fedak PWM, Stephens EH, Gleason TG, Girdauskas E, Ikonomidis JS, et al. The American association for thoracic surgery consensus guidelines on bicuspid aortic valve-related aortopathy: executive summary. *J Thorac Cardiovasc Surg*. (2018) 156:473–80. doi: 10.1016/j.jtcvs.2017.10.161

4. Prakash SK, Michelena HI, Milewicz DM. Bicuspid aortic valve and thoracic aortic disease: further evidence of clinically silent but deadly risk to family members of affected individuals. *Circulation*. (2023) 148:648–50. doi: 10.1161/CIRCULATIONAHA.123.065406

5. Tzemos N, Therrien J, Yip J, Thanassoulis G, Tremblay S, Jamorski MT, et al. Outcomes in adults with bicuspid aortic valves. *JAMA*. (2008) 300:1317–25. doi: 10.1001/jama.300.11.1317

6. Simpson JM, Pushparajah K. Dilatation of the aorta in bicuspid aortic valve disease. *Circ Cardiovasc Imaging*. (2020) 13:e010448. doi: 10.1161/CIRCIMAGING.120.010448

7. Kim HH, Lee S, Lee SH, Youn YN, Yoo KJ, Joo HC. The long-term fate of ascending aorta aneurysm after wrapping versus replacement. *J Thorac Cardiovasc Surg*. (2022) 164:463–74.e4. doi: 10.1016/j.jtcvs.2020.12.110

8. Winkler A, Puiu P, Krombholz-Reindl P, Votsch A, Steindl J, Neuner M, et al. Impact of concomitant replacement of the ascending aorta in patients undergoing aortic valve replacement on operative morbidity and mortality. *Eur J Cardiothorac Surg*. (2022) 61:587–93. doi: 10.1093/ejcts/ezab420

9. Idrees JJ, Roselli EE, Blackstone EH, Lowry AM, Soltesz EG, Johnston DR, et al. Risk of adding prophylactic aorta replacement to a cardiac operation. *J Thorac Cardiovasc Surg*. (2020) 159:1669–78.e10. doi: 10.1016/j.jtcvs.2019.05.001

10. Girdauskas E, Borger MA, Secknus MA, Girdauskas G, Kuntze T. Is aortopathy in bicuspid aortic valve disease a congenital defect or a result of abnormal hemodynamics? A critical reappraisal of a one-sided argument. *Eur J Cardiothorac Surg*. (2011) 39:809–14. doi: 10.1016/j.ejcts.2011.01.001

11. Nightingale M, Guzzardi DG, Barker AJ, Malaisrie SC, McCarthy PM, Markl M, et al. Elastin integrity in bicuspid valve-associated aortopathy is associated with altered biomechanical properties and influenced by age. *Ann Cardiothorac Surg*. (2022) 11:426–35. doi: 10.21037/acs-2022-bav-17

12. Isselbacher EM, Preventza O, Hamilton Black J 3rd, Augoustides JG, Beck AW, Bolen MA, et al. 2022 ACC/AHA guideline for the diagnosis and management of aortic disease: a report of the American heart association/American college of cardiology joint committee on clinical practice guidelines. *Circulation*. (2022) 146:e334–482. doi: 10.1161/CIR.0000000000001106

13. Kallenbach K, Sundt TM, Marwick TH. Aortic surgery for ascending aortic aneurysms under 5.0 cm in diameter in the presence of bicuspid aortic valve. *JACC Cardiovasc Imaging*. (2013) 6:1321–6. doi: 10.1016/j.jcmg.2013.08.009

14. Debl K, Djavidani B, Buchner S, Poschenrieder F, Schmid FX, Kobuch R, et al. Dilatation of the ascending aorta in bicuspid aortic valve disease: a magnetic resonance imaging study. *Clin Res Cardiol*. (2009) 98:114–20. doi: 10.1007/s00392-008-0731-0

15. Bauer M, Pasic M, Meyer R, Goetze N, Bauer U, Siniawski H, et al. Morphometric analysis of aortic media in patients with bicuspid and tricuspid aortic valve. *Ann Thorac Surg*. (2002) 74:58–62. doi: 10.1016/S0003-4975(02)03650-0

16. Girdauskas E, Disha K, Borger MA, Kuntze T. Long-term prognosis of ascending aortic aneurysm after aortic valve replacement for bicuspid versus tricuspid aortic valve stenosis. *J Thorac Cardiovasc Surg*. (2014) 147:276–82. doi: 10.1016/j.jtcvs.2012.11.004

17. Hardikar AA, Marwick TH. Surgical thresholds for bicuspid aortic valve associated aortopathy. *JACC Cardiovasc Imaging*. (2013) 6:1311–20. doi: 10.1016/j.jcmg.2013.10.005

18. Liu S, Shi Y, Liu R, Tong M, Luo X, Xu J. Early prognosis of reduction ascending aortoplasty in patients with aortic valve disease: a single Center's experience. *Ann Thorac Surg*. (2017) 103:511–6. doi: 10.1016/j.athoracsur.2016.06.005

19. Polvani G, Barili F, Dainese L, Topkara VK, Cheema FH, Penza E, et al. Reduction ascending aortoplasty: midterm follow-up and predictors of redilatation. *Ann Thorac Surg*. (2006) 82:586–91. doi: 10.1016/j.athoracsur.2006.03.025

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TYPE Case Report
PUBLISHED 06 January 2025
DOI 10.3389/fcvm.2024.1480392

OPEN ACCESS

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RECEIVED 13 August 2024
ACCEPTED 26 November 2024
PUBLISHED 06 January 2025

CITATION
Rodgers J, Hill M and Zeigler S (2025) Case Report: *Fusarium falciforme* pericardial and sternal wound infection following orthotopic heart transplantation.
Front. Cardiovasc. Med. 11:1480392.
doi: 10.3389/fcvm.2024.1480392

Case Report: *Fusarium falciforme* pericardial and sternal wound infection following orthotopic heart transplantation

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Fusarium, a genus of soil and vegetation-based fungi, is a rare cause of infections in immunocompromised individuals, including transplant recipients. In this case, we describe successful treatment of *Fusarium falciforme* mediastinitis in the recipient of an orthotopic heart transplant. Treatment included multiple courses of combination antibiotic and antifungal therapy several surgical debridements, continuous mediastinal irrigation with antifungal agents, and staged closure with an omental flap. This is the first report describing successful eradication of *fusarium sp.* mediastinitis and provides a template for treating complex cases of mediastinitis and osteomyelitis.

KEYWORDS

heart transplant, fungal infection, mediastinitis, infection, transplant, wound vac, wound vac therapy

Introduction

Fusarium species are soil and vegetation-based fungi that can cause infections in immunocompromised individuals (1). This includes recent solid organ transplant recipients who are placed on immunosuppression, posing a unique challenge in postoperative management (1). Once diagnosed, invasive fungal infections are associated with an approximately 60% overall case-fatality rate among solid organ transplant recipients (2). Here, we present a case of successfully treated pericarditis and osteomyelitis caused by *Fusarium falciforme* in a patient following orthotopic heart transplant. To our knowledge, this is the first reported case in which sternal and pericardial infection with this organism was successfully treated.

Case description

A 54-year-old male with end-stage heart failure with a past medical history of type 2 diabetes (HbA1c 6.5), sarcoidosis with cardiomyopathy, ventricular tachycardia, and hypertension presented to our institution with worsening fatigue secondary to exacerbation of preexisting heart failure. Prior interventions included cardiac resynchronization therapy with cardioverter defibrillator implantation. Five days after admission, he was transferred to the cardiac ICU due to a decline in hemodynamic status. Inotropic therapy was initiated, and his transplant listing status was upgraded to 1A, meaning he was highest priority for receiving a donor heart.

Six weeks after admission, during which time the patient underwent optimization with milrinone and diuretic therapy, a donor heart became available. Bacterial culture from the

donor heart did not grow any microorganisms. During the perioperative course, the patient was treated with nystatin 500,000 units mouthwash four times per day and fluconazole 200 mg IV every 48 h. Post-operatively, the patient was placed on an immunosuppressive regimen including mycophenolate, tacrolimus, and methylprednisolone.

On postoperative day (POD) 10, he developed significant leukocytosis and was treated empirically with broad spectrum antibiotics and micafungin 100 mg IV once daily for 3 days. On POD 11, the patient was found to have a large pericardial effusion requiring pericardial window and drainage. Cultures of the pericardial fluid grew *Fusarium falciforme*, as identified through combined phenotypic characterization and DNA sequencing.

In collaboration with the Infectious Disease service, the patient was started on voriconazole 350 mg IV twice daily and amphotericin B 650 mg IV daily. Two weeks following these recommendations, the strain of *Fusarium* was determined to be resistant to voriconazole. Amphotericin B was added for its suggested synergy with voriconazole (3). Prolonged use of amphotericin B led to acute kidney injury requiring intermittent dialysis. This ultimately progressed to chronic renal failure.

With this treatment, blood cultures remained negative. Five weeks after transplantation, the patient was discharged on IV amphotericin B 650 mg daily monotherapy through a peripherally-inserted central catheter (PICC). Immune suppression consisted of oral mycophenolate 500 mg four times daily and tacrolimus 0.5 mg twice daily.

Six days after discharge, the patient was readmitted with nausea, vomiting, and fever. Admission labs were notable for leukocytosis and imaging revealed dehiscence of the chest wall incision with inflammatory changes and gas surrounding the sternotomy concerning for deep sternal wound infection and osteomyelitis (Figure 1). The patient was promptly restarted on IV voriconazole 500 mg every 12 h twice as a loading dose followed by 300 mg every 12 h for maintenance dosing.

The patient was taken to the operating room urgently. Loculated, turbid, foul-smelling fluid was encountered in the pericardial space. All soft loculations were debrided to completely mobilize the heart. Superficial soft tissue and the sternal edges were debrided with curets. Osteomyelitis appeared limited. The cavity was then irrigated with voriconazole irrigation followed by pulse lavage with voriconazole irrigation.

A continuous irrigation system and negative pressure therapy system was then constructed, as demonstrated in Figure 2. This was designed to allow antimicrobial solution to percolate from the posterior mediastinum anteriorly through the sternotomy and out the dressing. A 19 Fr Blake drain was placed in the posterior pericardium, tunneling it through the subcutaneous tissue and securing it in the usual manner. The wound was irrigated with voriconazole solution through the drain. Negative pressure wound vacuum sponges were then placed in the anterior mediastinum and between the sternal edges. The wound vacuum was initially irrigated every 12 h with amphotericin B solution. However, later postoperative cultures from this initial debridement grew methicillin-resistant *Staphylococcus aureus* (MRSA). This prompted the alternation of Adam's solution containing bacitracin, vancomycin, and gentamicin in addition to amphotericin B for the known *Fusarium* infection. Two consecutive debridements were performed three and four days after admission, respectively. During each of these two washouts, the mediastinum had no evidence of purulence or uncontrolled infection. Posterior irrigation and anterior vacuum suction was continued.

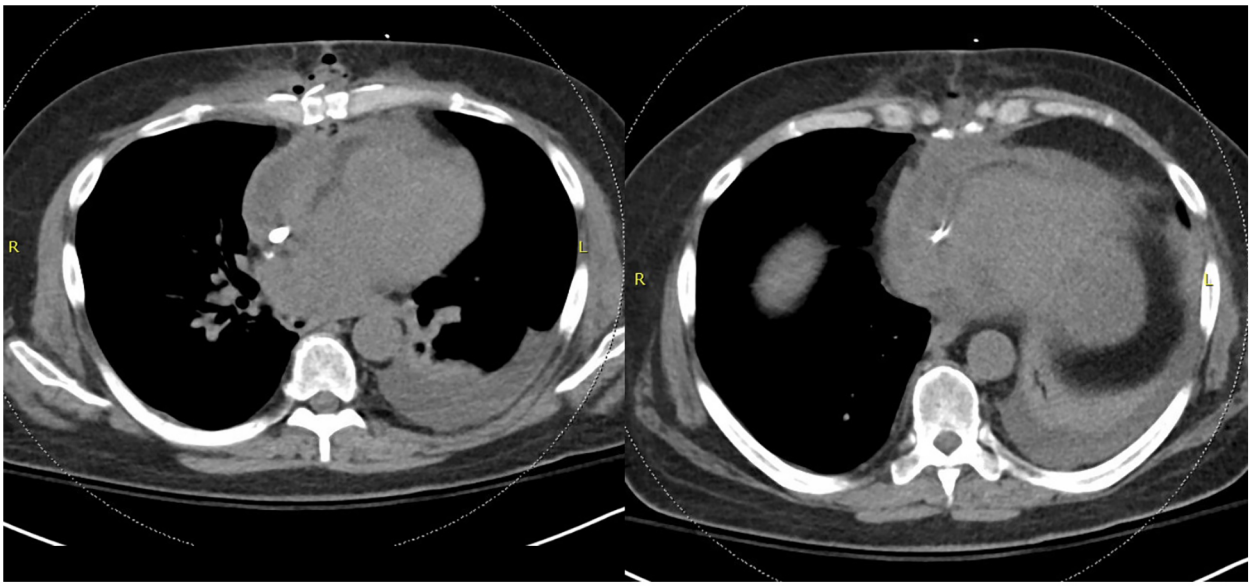

FIGURE 1
Non-contrasted CT chest demonstrating dehiscence of the chest wall incision with inflammatory changes and gas concerning for deep sternal wound infection.

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FIGURE 2
An example setup of the continuous wound irrigation system. A 19 Fr Blake drain is placed in the posterior pericardium and tunneled through the subcutaneous tissue before being secured in the usual manner. This setup is proceeded by the placement of wound vacuum sponges in the anterior mediastinum, followed by sternal closure. Voriconazole solution is flushed through the drain.

We then planned for definitive closure using an omental flap. Eleven days after admission, the patient underwent laparoscopic omental flap harvest. The sternal wound vac was removed and the wound was explored and debrided. The flap was then passed through a cruciate incision, two fingerbreadths in width, in the diaphragm and tunneled into the pericardial space. Bilateral pectoralis advancement flaps were then harvested. The omental flap was tacked to cover the aorta and right atrium. 19 Fr Blake drains were placed beneath each myocutaneous pectoralis flap, and the flaps were then advanced to the midline and closed over the omental flap. The superficial closure was buttressed with retention sutures. The sternum was left in non-union. An incisional vacuum dressing was placed.

The patient was treated with 8 weeks of IV vancomycin for MRSA mediastinitis and bacteremia, and 6 months IV amphotericin B for fungal mediastinitis. His renal failure persisted and progressed to ESRD. He required a PEG tube for nutrition. Eight weeks from his admission, the patient was discharged to a subacute rehabilitation facility with a PICC line in place. Blood cultures remained negative. Beta-D glucan levels remained elevated for 6 months after his re-exploration and were normal 18 months postoperatively.

The patient survived 5 years since heart transplant. He remained without severe primary graft dysfunction with an ejection fraction >55% without RV or LV dysfunction. He had no recurrence of bloodstream infection until he died. At that time, he was preparing to be listed for kidney transplantation, and developed recurrent MRSA bacteremia related to his dialysis access.

Discussion

Fusarium falciforme is a ubiquitous species of *Fusarium* that exists in the air, soil, water, and vegetation (1). Localized infections can occur in both immunocompetent and immunocompromised individuals, and normally first noticed by a dermatologic presentation (1, 4). They can also be diagnosed radiographically with CT imaging or 18-FDG PET/CT imaging. *Fusarium* has also been shown to be a rare agent of disseminated infection in immunocompromised individuals, which has been reported in patients in treatment for acute myeloid leukemia and other individuals with profound neutropenia (1, 5). However, fusariosis is only rarely associated with recent solid organ transplantation, including orthotopic heart transplantation (4). This patient developed a localized fusarium infection that was also associated with MRSA wound infection and bacteremia.

Data regarding the treatment of disseminated and visceral *Fusarium* infection is somewhat lacking because of the low occurrence of disseminated *Fusarium* infections with successful treatment. Previous studies suggest current first-line treatment for disseminated fusariosis with either voriconazole, amphotericin B, or liposomal amphotericin B (4). However, other studies have noted the potential limitation of using amphotericin B monotherapy in the eradication of this fungal agent (6). Studies looking at combination therapy for this disease is even more limited (1). For this unique case, it was decided to treat his fungal pericarditis and osteomyelitis with a combination of amphotericin B and voriconazole, as this had been reported to be previously successful in a case of *Fusarium* infection causing endocarditis (7). Though sensitivity studies determined that this strain of *Fusarium* had resistance to voriconazole alone, it was continued as part of the therapeutic regimen because of the previously reported synergy among amphotericin B- and voriconazole-based therapies, respectively (3).

To our knowledge, this is the first report of a continuous irrigation system containing an antifungal solution for a patient with fungal mediastinitis. Previous studies have reported thoracic continuous saline irrigation systems have been constructed for the purpose of descending necrotizing mediastinitis (8). In this study, nonantibiotic saline irrigation was constructed in addition to standard-of-care IV antibiotics. Even so, continuous saline irrigation resulted in significantly decreased length of hospital stay and a significantly shorter drainage period than patients with necrotizing mediastinitis who received the current standard of care. Another previous case report demonstrates the efficacy of continuous antifungal irrigation in a case of renal infection (9). In our patient, we decided to apply this continuous irrigation to fungal mediastinitis by adding voriconazole and later, amphotericin B to the saline irrigation solution.

Another unique aspect of the treatment of our case was the use of an omental flap for the use of fungal mediastinitis. This two-step omental flap creation was described previously for the successful

resolution of Candidal aortic graft infection and omental flaps have been used in a variety of thoracic defects, including sternal wound infections, bronchopleural fistulas, chest wall defects, and other reconstructive operations (10). As the authors noted, an omental flap absorbs infectious secretions while also providing a surface of increased vascularization for antibiotic delivery (10). Similarly to the patient described in the previous case, this mediastinal omental flap allowed for recovery after the continuous antifungal irrigation solution.

Conclusions

In conclusion, here we describe a unique case of an orthotopic heart transplant complicated by mediastinal *Fusarium falciforme*. The patient was successfully treated with a combination of antibiotic therapy, surgical debridements with continuous antifungal irrigation system installation, and eventual closure with mediastinal omental flap insertion. Overall, we demonstrate the successful combination of multiple treatment modalities in individuals with rare post-transplantation fungal mediastinal infections.

Data availability statement

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

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

JR: Investigation, Writing – original draft, Writing – review & editing. MH: Data curation, Investigation, Writing – original draft, Writing – review & editing. SZ: Conceptualization, Investigation, Supervision, Writing – review & editing.

Funding

The author(s) declare that no financial support was received for the research, authorship, and/or publication of this article.

Acknowledgments

We would like to thank the family members of the patient discussed in this report for granting us permission to present this story. We would also like to thank Megan McNicholas, a medical student who aided the treatment team and began the collection of information for the writing of this case report.

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

References

1. Nucci M, Anaissie E. Fusarium infections in immunocompromised patients. *Clin Microbiol Rev*. (2007) 20(4):695–704. doi: 10.1128/CMR.00014-07

2. Bodro M, Sabe N, Comila A, Ayats J, Baliellas C, Roca J, et al. Risk factors, clinical characteristics, and outcomes of invasive fungal infections in solid organ transplant recipients. *Transplant Proc*. (2012) 44(9):2682–5. doi: 10.1016/j.transproceed.2012.09.059

3. Siopi M, Siafakas N, Vourli S, Zerva L, Meletiadis J. Optimization of polyene-azole combination therapy against aspergillosis using an *in vitro* pharmacokinetic-pharmacodynamic model. *Antimicrob Agents Chemother*. (2015) 59(7):3973–83. doi: 10.1128/AAC.05035-14

4. Sampathkumar P, Paya CV. Fusarium infection after solid-organ transplantation. *Clin Infect Dis*. (2001) 32(8):1237–40. doi: 10.1086/319753/2/32-8-1237-TBL002

5. Yun SJ, Shin MG, Choi C, Kim HJ, Lee JB, Kim SJ, et al. Fatal disseminated angioinvasive Fusarium falciforme infection in a patient with acute myeloid leukaemia. *Br J Dermatol*. (2007) 157(2):407–9. doi: 10.1111/J.1365-2133.2007.08020.X

6. Nambiar P, Cober E, Johnson L, Brizendine KD. Fatal Fusarium infection manifesting as osteomyelitis following previous treatment with amphotericin B in a multi-visceral transplant: case report and review of Fusarium infections in solid organ transplantation. *Transpl Infect Dis*. (2018) 20(3):e12872. doi: 10.1111/TID.12872

7. Guzman-Cottrill JA, Zheng X, Chadwick EG. Fusarium solani endocarditis successfully treated with liposomal amphotericin B and voriconazole. *Pediatr Infect Dis J*. (2004) 23(11):1059–61. doi: 10.1097/01.INF.0000143649.90952.41

8. Ohashi T, Kawago M, Hirai Y, Yata Y, Fusamoto A, Iguchi H, et al. Efficacy of continuous saline irrigation therapy for descending necrotizing mediastinitis. *Surg J*. (2023) 09(03):e107–11. doi: 10.1055/s-0043-1775559

9. van Merode NAM, Pat JJ, Wolfhagen MJHM, Dijkstra GA. Successfully treated bilateral renal fungal balls with continuous Anidulafulgin irrigation. *Urol Case Rep*. (2021) 34:101468. doi: 10.1016/J.EUCR.2020.101468

10. Andrade D, Vinck E E, Niño Torres L. Two-stage omental flap approach for ascending aortic graft infection. *Braz J Cardiovasc Surg*. (2020) 35(3). doi: 10.21470/1678-9741-2020-0040

frontiers | Frontiers in Cardiovascular Medicine

TYPE Case Report
PUBLISHED 31 January 2025
DOI 10.3389/fcvm.2025.1463764

OPEN ACCESS

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RECEIVED 12 July 2024
ACCEPTED 09 January 2025
PUBLISHED 31 January 2025

CITATION
Xia J, Qiu Y, Chang S, Feng Y, Zhang H and Wang X (2025) Case Report: Surgery combined with extracorporeal membraneoxygenation for acute type A aortic dissection complicated with acute myocardial infarction.
Front. Cardiovasc. Med. 12:1463764.
doi: 10.3389/fcvm.2025.1463764

Case Report: Surgery combined with extracorporeal membraneoxygenation for acute type A aortic dissection complicated with acute myocardial infarction

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Background: Acute myocardial infarction (AMI) is one of the most serious complications of acute type A aortic dissection (ATAAD) and markedly increases patient mortality. Simultaneous treatment strategies remain controversial. How to improve the treatment of these patients remains a critical challenge for cardiovascular surgeons.
Case presentation: All three patients who experienced chest pain were admitted to emergency department of our hospital. The 12-lead electrocardiogram revealed ST-segment depression, myocardial enzyme levels were significantly elevated. Emergency physicians diagnosed ATAAD with AMI, and emergency surgery was planned in collaboration with the cardiovascular surgery team. One-stage surgery for coronary revascularization and central aortic repair were performed, extracorporeal membrane oxygenation (ECMO) was implanted, ECMO was discontinued when hemodynamics stabilized. The patient ultimately recovered well and was discharged.
Conclusion: For ATAAD combined with right ventricular AMI, one-stage surgery for coronary revascularization and central aortic repair, supported by ECMO as bridge, can be life-saving treatment strategy, the prognosis for all three patients was excellent.

KEYWORDS

acute myocardial infarction (AMI), acute type A aortic dissection (ATAAD), extracorporeal membrane oxygenation (ECMO), one-stage surgery, malperfusion syndrome

Abbreviations

AMI, acute myocardial infarction; ATAAD, acute type A aortic dissection; CABG, coronary artery bypass grafting; CPB, cardiopulmonary bypass; CTA, computed tomography angiogram; ECMO, extracorporeal membrane oxygenation; IABP, intra-aortic balloon pump; IRAD, internation registry of acute aortic dissection; LAD, left anterior descending artery; LVEF, left ventricular ejection fraction; MACCE, major adverse cardiac and cerebrovascular events; MPS, malperfusion syndrome; NSTEMI, non- St-segment elevation myocardial infarction; PCI, percutaneous coronary intervention; PDA, posterior descending branch; PL, left ventricular posterior branch; RCA, right coronary artery; TTFM, transit time flow measurement.

10.3389/fcvm.2025.1463764

Introduction

Acute type A aortic dissection (ATAAD) is a highly lethal cardiovascular disease, with the first 24–48 h being a critical time window for mortality (1). Advances in medical technology have led to a gradual decrease in the mortality rate of ATAAD. Notably, based on data from the International Registry of Acute Aortic Dissection (IRAD), Harris et al. (2) found that the 48-hour mortality rate decreased to 23.7% (0.5%/hour) following drug treatment but to 4.4% (0.09%/hour) following surgical treatment. Our strategy for treating ATAAD and reducing mortality emphasizes early diagnosis and immediate surgical treatment. Malperfusion syndrome (MPS) is a severe complication of ATAAD, characterized by high mortality and poor clinical outcomes despite advancements in diagnosis and treatment strategies (3). ATAAD combined with myocardial infarction (AMI) represents one of the most serious forms of MPS (4). Currently, no treatment guidelines have been established for ATAAD combined with AMI, and treatment strategies remain controversial, with successful case outcomes primarily documented in case reports. In recent years, we have successfully treated three cases of ATAAD complicated with AMI using surgery and extracorporeal membrane oxygenation (ECMO) in combination, as shown in Table 1. This summary aims to enhance our understanding and management of this complex condition.

TABLE 1 Baseline characteristics of ATAAD complicated with AMI.

	Case 1	Case 2	Case 3
Age (years)	44	41	58
Sex (Male/female)	Male	Male	Male
Height (cm)	168	170	170
Weight (Kg)	88.5	49	68
Classification of acute myocardial infarction	Non- ST-segment elevation myocardial infarction (NSTEMI)	NSTEMI	NSTEMI
Aortic sinus diameter (mm)	57	50	57
Ascending aortic diameter (mm)	50	50	41
Left ventricular end diastolic diameter (mm)	40	51	66
Ejection fraction (%)	59	58	42
Platelet ($\times 10^9$)	152	115	160
Creatinine (μmol/L)	107	66.3	122
Disease onset-operation time	27	65	28
Classification of Neri	B	B	C
Drainage fluid on first day after surgery (ml)	750	870	910
Input red blood cells (u)	11.5	12u	10u
Input plasma (ml)	2,200	1,400	400
Input platelets (u)	2	4u	2u
ECMO time (h)	83	97	106
IABP time (h)		168	
CRRT time (h)			504
Ventilator time(h)	136	213	221
ICU time (h)	205	430	677
Hospital stays (h)	589	982	677

Case reports

Case 1

A 44-year-old man (height 168 cm, weight 88.5 kg) was admitted to the emergency department of our hospital with chest pain persisting for 8 h. He had a history of hypertension but did not take medication regularly. His vital signs upon admission included a right arm blood pressure of 138/77 mmHg, a pulse of 70 beats/min, a respiratory rate of 18 breaths/min, and an oxygen saturation (SpO2) of 97%. A 12-lead electrocardiogram revealed ST-segment depression in the II, III, and aVF leads. Myocardial enzyme levels were significantly elevated, including CK-MB at 182 ng/ml (0–6 ng/ml) and troponin at 24.54 ng/ml (0–0.08 ng/ml). A pre-operative computed tomography angiogram (CTA) of the entire aorta, coronary artery and supra-aortic vessels revealed ATAAD with a rupture located in the ascending aorta (Figure 1A–B). The dissection flap extended from the ascending aorta to bilateral common iliac artery, right external iliac artery and left internal iliac artery. Celiac trunk artery, superior mesenteric artery, bilateral renal artery and most of intercostal arteries originate from the true lumen, while the inferior mesenteric artery originate from the false lumen. Eccentric thickening of the wall of the proximal segment of the right coronary artery and the proximal segment of the left ventricular posterior branch was observed. A small aneurysm was noted in the M2 segment of the left middle cerebral artery, and the lumen of the right anterior cerebral artery appeared slender. Pre-operative echocardiography revealed coordinated movement of the left and right ventricles with normal contraction amplitude and a left ventricular ejection fraction (LVEF) of 59%. Emergency physicians diagnosed ATAAD with AMI, and emergency surgery was planned in collaboration with the cardiovascular surgery team. For the surgery performed via median sternotomy, cardiopulmonary bypass (CPB) was established through the axillary artery, femoral artery, and right atrium. The ascending aorta was clamped proximal to the brachiocephalic artery and opened to inspect the coronary ostial lesions. A cold blood cardioplegic solution was perfused through the coronary ostium, and no intimal tear was found. The heart was covered with ice. The dissected aorta was resected, intima and media of the aorta were resected, the false lumen blood clots were cleared, and a large epicardial hematoma on the right ventricular surface was explored. Sequential coronary artery bypass grafting (CABG) was performed using a saphenous vein graft from the left ventricular posterior branch (PL) to the posterior descending branch (PDA). Proximal aortic reconstruction was performed using the adventitial inversion technique. Circulatory arrest was initiated at a bladder temperature of 28°C with bilateral-anterograde cerebral perfusion. A 30-mm self-expandable elephant trunk stent graft was inserted into the true lumen of the descending aorta, and aortic arch

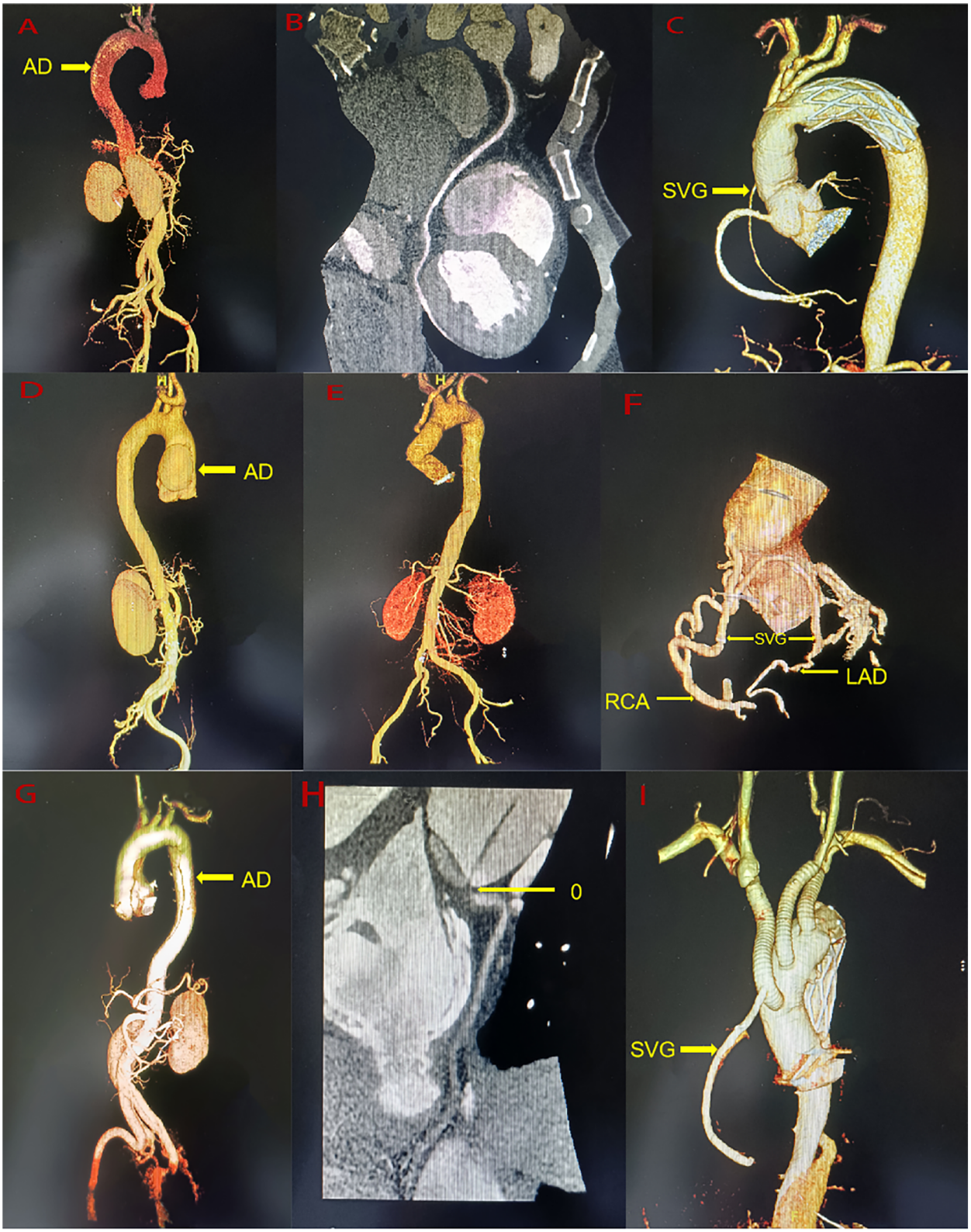

FIGURE 1
(A) A pre-operative the entire aorta CTA of case 1. **(B)** A pre-operative the right coronary artery CTA of Case 1. **(C)** A post-operative the entire aorta CTA of Case 1. **(D)** A pre-operative the entire aorta CTA of Case 2. **(E)** A post-operative the entire aorta CTA of Case 2. **(F)** A post-operative the coronary artery CTA of Case 2. **(G)** A pre-operative the entire aorta CTA of Case 3. **(H)** A pre-operative the right coronary artery CTA of Case 3. **(I)** A post-operative the entire aorta CTA of Case 3. AD, aortic dissection; RAC, right coronary artery; LAD, anterior descending branch; SVG, saphenous vein graft.

reconstruction was performed using a 30-mm four-branched arch graft. When the heart resumed beating and body temperature normalized, intraoperative transesophageal echocardiography revealed decreased left heart function with an LVEF of 32%, the patients have evidence of cardiogenic shock (systemic systolic pressure <90, urine output <30 ml/hour, lactate >2), necessitating the initiation of ECMO through the axillary artery and femoral vein (rotation speed: 2,500 R/min, flow rate: 3.360 L/min). The right coronary saphenous vein graft flow was 58 ml/min, and the pulsatility index was 2.0 by transit time flow measurement (TTFM). The CPB duration was 337 min, aortic cross-clamping time was 153 min, and the circulatory arrest time was 19 min. ECMO was discontinued on post-operative day 3, and the patient was extubated on day 7. The patient spent 18 days in intensive care and 24 days in the hospital. Echocardiography at discharge revealed hypokinesis of the middle and the lower segments of the right ventricle and lower posterior wall of the left ventricle, with an LVEF of 55%. Ultrasound at discharge revealed venous thrombosis in the bilateral lower limbs and right upper limbs. The CTA revealed bilateral scattered pulmonary embolism with an unobstructed artificial vessel and a well-dilated elephant trunk stent without internal leakage. The false lumens distal to the stent had thrombosed. At 2 years and 6 months of follow-up, the patient exhibited normal heart function and could perform normal daily activities. Recent echocardiography revealed normal left and right ventricular wall movements, with an LVEF of 63%. Recent CTA revealed an unobstructed great saphenous vein graft, occlusion of the left ventricular posterior branch, unobstructed artificial vessels, a well-attached elephant trunk stent to the aortic wall, thrombosis and occlusion of the false lumen (Figure 1C).

Case 2

A 41-year-old man (height 170 cm, weight 49 kg) was admitted to the emergency department of our hospital with persistent chest pain for 2 days. He had no prior medical history. His vital signs upon admission included a right arm blood pressure of 122/75 mmHg, a pulse of 99 beats/min, a respiratory rate of 17 breaths/min, and a SpO2 of 97%. A 12-lead electrocardiogram revealed ST-segment depression in leads V4-V6. Laboratory tests showed elevated troponin at 2.54 ng/ml (0–0.08 ng/ml) and CK-MB at 6.76 ng/ml (0–6 ng/ml). Pre-operative CTA of the entire aorta, coronary artery and supra-aortic vessels revealed ATAAD, with the entry located in the ascending aorta (Figure 1D). The dissection was limited from the aortic sinus to ascending aorta, with a penetrating ulcer in the initial segment of the left carotid artery, severe stenosis of celiac trunk artery, but no significant stenosis in the coronary arteries. Pre-operative echocardiography revealed mild-to-moderate aortic valve regurgitation and normal ventricular wall movement, with an LVEF of 58%. Emergency physicians diagnosed type A AD with AMI, with surgery arranged after discussion with the Cardiovascular Surgery team. The surgery was performed via median sternotomy, which included establishing CPB through the axillary artery and right atrium. The ascending aorta was clamped proximal to the brachiocephalic artery and opened to inspect the coronary ostial lesions. Cold del Nido cardioplegic solution was administered through the coronary ostium with no intimal tear observed. The heart was covered in ice, and the intima and media of the aortic dissection were resected. The right-non-junction of the aortic valve was repaired with a 5-0 suture. Aortic sinus reconstruction was conducted using the adventitial inversion technique and bovine pericardium. Replacement of the ascending aorta was performed using a 30-mm straight vessel. The left carotid artery was clamped, and the penetrating ulcer was resected and replaced with an 8-mm straight graft. After the heart resumed beating, the aortic sinus dissection progressed, causing a hematoma that compressed the left and right coronary arteries. Esophageal ultrasound revealed obstruction on coronary blood flow and moderate aortic valve regurgitation. The ascending aorta was reclamped, and the anastomotic site was opened. Cold del Nido cardioplegic solution was directly perfused anterogradely from the coronary ostial. A thorough resection of the sinus dissection tissue was performed, the aortic valve was removed, and Bentall surgery was conducted. After the heart resumed beating, the movement of the right and left ventricular anterior walls was weakened. CABG was performed using a saphenous vein for the left anterior descending artery (LAD) and right coronary artery (RCA) during CPB. Intraoperative transesophageal echocardiography revealed decreased left and right heart function with an LVEF of 30%. The patients have evidence of cardiogenic shock (systemic systolic pressure <90, urine output <30 ml/hour) until after an intra-aortic balloon pump (IABP) was inserted, necessitating the initiation of ECMO through the femoral artery and femoral vein to unload the heart and restore cardiac function (rotation speed: 2,100 R/min, flow rate: 3.0 L/min). The RCA-saphenous vein graft flow was 72 ml/min, and the pulsatility index was 2.1 by TTFM. The LAD-saphenous vein graft flow was 58 ml/min, and the pulsatility index was 2.0 by TTFM. The CPB duration was 768 min, and the aortic cross-clamping time was 182 min. ECMO was discontinued on post-operative day 4. IABP was removed on post-operative day 7. The patient was extubated after 8 days and 21 h. The patient spent 17 days in intensive care and 40 days in the hospital. Echocardiography at discharge revealed normal left and right ventricular wall movement, with an LVEF of 60%. A CTA at discharge revealed an unobstructed ascending aorta and an artificial left carotid artery blood vessel (Figure 1E–F). At the 2 years and 4 -month follow-ups, the patient exhibited normal heart function and could perform normal daily activities. Recent echocardiography revealed normal left and right ventricular wall movement, with an LVEF of 58%. Recently, CTA revealed an unobstructed saphenous vein vessel, 50%–70% stenosis in the proximal right coronary artery, irregular and mild stenosis in the left main artery, unobstructed artificial blood vessels in the ascending aorta, and an artificial left carotid artery blood vessel.

Case 3

A 58-year-old man (height 170 cm, weight 68 kg) was admitted to emergency department of our hospital after experiencing chest

pain for 22 h. His vital signs upon admission included right arm blood pressure of 87/51 mmHg, a pulse rate of 89 beats/min, a respiratory rate of 17 breaths/min, and a SpO2 of 98%. A 12-lead electrocardiogram revealed ST-segment depression in the I and aVL leads and pathological Q waves in II, III, and aVF leads. Myocardial enzyme levels were significantly elevated, with CK-MB at 142 ng/ml (0–5.85 ng/ml) and troponin exceeding 3 ng/ml (0–0.018 ng/ml). The Pre-operative CTA of the entire aorta, coronary artery and supra-aortic vessels showed ATAAD with the rupture located in the ascending aorta, dissection flap extending from the root of the aorta to both internal and external iliac artery, aortic sinus aneurysm, and occlusion of the right coronary ostial (Figure 1G–H). Superior mesenteric artery, right renal artery, the inferior mesenteric artery and most of intercostal arteries originate from the true lumen, while celiac trunk artery and left renal artery originate from the false lumen. Pre-operative echocardiography revealed severe aortic regurgitation, with an LVEF of 42%. Emergency physicians diagnosed ATAAD with AMI, with emergency surgery arranged after discussion with the Cardiovascular Surgery team. The surgery performed in median sternotomy, involved establishing CPB through femoral artery and right atrium. The ascending aorta was clamped proximal to the brachiocephalic artery and opened to inspect the coronary ostial lesions. The left coronary ostial was normal, but the right coronary ostial displayed an intima tear located beside the right coronary artery ostium but not inside the right coronary artery ostium. Cold blood cardioplegic solution was perfused through the coronary ostium, and the heart was covered with ice. CABG was performed using a saphenous vein for the RCA, and the right coronary artery ostial was closed with retrograde cold blood cardioplegic solution through the saphenous vein. The aortic valve was resected, and Bentall surgery was performed. Bilateral cerebral perfusion was established through the right innominate artery and left carotid artery. Circulation arrest was induced when the bladder temperature dropped to 28 °C, combination with bilateral anterograde cerebral perfusion. A 24-mm self-expandable elephant trunk stent graft was inserted into the true lumen of the descending aorta, and aortic arch reconstruction was performed using a 26-mm four-branched arch graft. When the heart resumed beating and body temperature returned to normal, Intraoperative transesophageal echocardiography revealed decreased left and right heart function with an LVEF of 35%, the patients have evidence of cardiogenic shock (systemic systolic pressure <90, urine output <30 ml/hour, lactate >2), necessitating the initiation of ECMO through the femoral artery and femoral vein (rotation speed: 2 200 R/min, flow rate: 3.2 L/min). The RCA-saphenous vein graft flow was 47 ml/min, the pulsatility index was 4.2 by TTFM. After returning to surgical intensive care unit (SICU), the patient exhibited no urine output and high lactate levels, requiring continuous renal replacement therapy (CRRT). The CPB duration was 307 min, aortic cross-clamping time was 184 min, and circulatory arrest time was 21 min. ECMO was discontinued on post-operative day 4 and the patient was extubated on day 9. The patient remained in the SICU for 28 days, before being transferred to the rehabilitation department. Echocardiography at discharge revealed a diffuse decline in left ventricular wall contraction and normal right ventricular wall movement, with an LVEF of 43%. Ultrasounds at discharge revealed right lower limb venous thrombosis. The CTA at discharge indicated unobstructed artificial and saphenous vein grafts, a well-expanded elephant trunk stent with no internal leakage, thrombosed false lumens distal to the stent, and a right upper lobe pulmonary embolism (Figure 1I). At 3-month follow-up, the patient showed a diffuse decline in heart function but was able to engage in normal daily activities. Recent echocardiography revealed a diffuse reduction in left ventricular wall contraction, while right ventricular wall movement remained normal, with an LVEF of 48%.

Discussion

The primary causes of mortality in ATAAD are aortic rupture and malperfusion (5). The objective of the surgery intervention is to excise the dissection in the ascending aorta and aortic arch to prevent aortic rupture and restore organ perfusion, thereby preventing necrosis. For uncomplicated ATAAD, timely central aortic repair is widely recognized as the most effective and safest treatment option. However, the occurrence of MPS markedly increases patient mortality and significantly worsens the prognosis (6, 7). Addressing how to improve the treatment of these patients remains a critical challenge for cardiovascular surgeons.

The incidence of ATAAD combined with AMI ranges from 5 to 7.1% (2, 8), and the associated mortality rate is extremely high (9). Currently, there is no large-scale clinical data available on the treatment outcomes for this condition, with surgical treatment only documented in a few cases (10). In the cases reported here, we optimized the surgical treatment strategy for ATAAD combined with AMI. Following coronary revascularization and central aortic repair, ECMO was employed as a "bridge-to-recovery" to help stabilize hemodynamics. The prognosis for all three patients was excellent. To date, only two relevant reports using this treatment approach have been published, as shown in Table 2 (11, 12).

AMI, due to the propagation of an aortic dissection into the coronary arterial wall or compression of the coronary arteries by a hematoma, is a critical and often fatal condition (13). The pathophysiological process involves bulging of the dissected false lumen at the branch orifice, leading to distal thrombosis, intimal detachment, and further extension of the dissection into the branch (14). In contrast to stable aortic dissection, AMI represents an immediate, life-threatening complication. The optimal treatment strategy remains controversial, with ongoing debate between one-stage and staged surgery. One-stage surgery can simultaneously address both aortic rupture and myocardial ischemia but is associated with a high surgical mortality rate (9). To mitigate surgical risks, a staged approach has been proposed, starting with percutaneous coronary intervention (PCI) bridging to restore coronary blood flow and heart function, followed by center aortic repair. However, this approach risks aortic rupture before definitive aortic repair (15, 16). Cardiovascular surgeons

TABLE 2 Summary of patients with ATAAD complicated with AMI treated by surgery combined with ECMO.

Reference	11	12	Case 1	Case 2	Case 3
Age (years)	36	42	44	41	58
Sex (Male/female)	Female	Male	Male	Male	Male
Diagnosis	ATAAD combined with AMI	ATAAD combined with AMI	ATAAD combined with AMI	ATAAD combined with AMI	ATAAD combined with AMI
Disease onset-operation time (hours)	144	unrecorded	27	65	28
Operation	Ascending aorta and total aortic arch replacement combined with elephant trunk stent implantation	Ascending aorta and total aortic arch replacement combined with elephant trunk stent implantation	Aortic sinus reconstruction + ascending aorta and total aortic arch replacement combined with elephant trunk stent implantation + CABG	Bentall + CABG	Bentall + total arch replacement and elephant trunk stent implantation + CABG
Coronary processing	True lumen of RCA not found; CABG halted	RCA recovery patency by PCI	Sequential CABG was performed from PL and PDA using saphenous vein graft	CABG was performed using saphenous vein for LAD and RCA	CABG was performed using saphenous vein for RCA
ECMO time (hours)	312	120	72	96	96
Normal daily activities	Yes	Yes	Yes	Yes	Yes
Follow-up time (months)	3	6	30	28	3

are thus confronted with difficult choices and challenges. Considering the specific characteristics of the lesions in the three patients, we carefully considered and optimized our treatment strategy.

We opted not to perform PCI for the three patients, instead choosing a one-stage surgical approach for coronary revascularization and central aortic repair. This was decided based on the following rationale: (1) Performing PCI in cases of ATAAD carries the risk of aortic rupture. Not all coronary artery involvements caused by ATAAD can be corrected with PCI; some require surgical repair of both the aorta and coronary arteries or simultaneous CABG. (2) Performing PCI requires the use of anticoagulants, increasing the risk of bleeding during surgery. There is also a significant risk of stent thrombosis post-surgery, often requiring CABG, thereby complicating the overall treatment. (3) The three patients, presenting with NSTEMI demonstrated relatively stable circulatory conditions, categorizing them as a low-risk population of acute ischemic events. (4) The patients primarily experienced right ventricular myocardial infarction. The right ventricle exhibits remarkable resilience to ischemic injury and a strong capacity for recovery even after prolonged occlusion. Consequently, the term "right ventricular infarction" is somewhat misleading, as an acutely ischemic right ventricle remains largely viable and robust (17). While right ventricular function can improve spontaneously without reperfusion, recovery can be slow and is often associated with significant in-hospital morbidity and mortality. Reperfusion accelerates the recovery of right ventricular function and improves overall clinical outcomes and survival rates. Additionally, using ECMO as a bridge can help reduce mortality and complications. Wang et al. (11) reported a case of complete right coronary artery occlusion associated with type A aortic dissection, where CABG was not performed, and right ventricular function gradually recovered postoperatively with ECMO support. Similarly, the three patients discussed experienced full recovery of their right heart function following surgery.

Neri et al. (18) introduced the famous Neri classification system based on the extension of the dissection. This system is both simple and practical for making decisions during surgery. According to this classification, the coronary ostia can be directly repaired for Neri A and several Neri B types, while coronary artery bypass grafting can be performed for certain Neri B types and Neri C types (19). Given preoperative myocardial injury and the potential for intraoperative cardiac arrest, myocardial protection during surgery is crucial. Therefore, exploration of coronary ostia lesions before cardioplegic perfusion is necessary to ensure effective myocardial protection during surgery (20). The cardioplegic solution is then perfused through the coronary ostium if no intimal tear is found. After performing CABG, retrograde perfusion is completed via a saphenous vein graft. If no intimal tear is identified, cardioplegic solution can be safely delivered through the coronary ostium. Following CABG, retrograde perfusion via a saphenous vein graft is completed to further optimize myocardial perfusion. In cases where anterograde perfusion through the coronary ostium is not feasible, direct perfusion into the coronary artery or retrograde delivery via the coronary sinus serve as effective alternatives. While coronary CTA is the primary imaging modality for preoperative coronary evaluation, its accuracy is lower than that of coronary angiography, so results should be interpreted with caution. During surgery, meticulous exploration of the coronary arteries is necessary to determine the need and precise location for CABG. In case 2, due to an inaccurate coronary CTA assessment and weakened heart function after reperfusion, an unplanned CABG was required.

Previous research has shown no difference in mortality between early CABG (within 72 h) and late CABG (beyond

72 h), with a similar incidence of major adverse cardiac and cerebrovascular events (MACCE) at 6-month follow-up among NSTEMI patients (21). IABP is a common device used to increase coronary blood flow. However, the IABP acute myocardial infarction complicated with cardiogenic shock, the IABP-SHOCK trial demonstrated that IABP cannot reduce all-cause mortality in AMI complicated by cardiogenic shock (22). As a result, current European guidelines have downgraded the recommendation for IABP use from class I C to class III B (23). Recently, the use of alternative devices such as Impella, TandemHeart, and ECMO has increased significantly. ECMO, which reduces cardiac preload and oxygen consumption while providing circulatory and respiratory support, has become our preferred and most critical assistive device for successfully weaning patients off CPB. In case 2, where there was no dissection in the descending aorta, both IABP and ECMO were used. In contrast, in Cases 1 and 3, where descending aortic dissection was present, only ECMO was employed.

We use heparin anticoagulant, an ACT range of 180–220s and an APTT range of 60–80s have been suggested for ECMO. Patients with type A dissection experience significant disruption in their coagulation function, which is further exacerbated by surgical trauma and the administration of large volumes of blood products. Consequently, post-surgery coagulation is often severely impaired. In addition, prolonged bed rest during recovery increases the risk of thrombosis. In the cases reported here, two developed pulmonary embolism, and two developed deep vein thrombosis in their limbs.

All three patients showed favorable recovery post-surgery, with follow-up periods ranging from 3 months to 2 years and 6 months. Postoperative evaluations indicated that both artificial vessels and saphenous vein grafts remained unobstructed, and right heart function had normalized, enabling patients to resume normal daily activities.

In conclusion, ATAAD combined with AMI is a rare and complex condition with no established international treatment guidelines. Surgical intervention is highly challenging, but with an experienced cardiovascular diagnosis and treatment team, one-stage surgery for coronary revascularization and central aortic repair, supported by ECMO as a bridge, can be life-saving.

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 participant/patient(s) for the publication of this case report.

Author contributions

JX: Writing – original draft. YQ: Resources, Writing – review & editing. SC: Resources, Writing – review & editing. YF: Data curation, Writing – review & editing. HZ: Writing – review & editing. XW: Writing – review & editing.

Funding

The author(s) declare financial support was received for the research, authorship, and/or publication of this article. This work was supported by the grants from the Open Project of the State Key Laboratory of Cardiovascular Diseases (2019kfyf-01), Key Technology Research and Device Development Project for Innovative Diagnosis and Treatment of Structural Heart Disease in the Southwest Plateau Region (202302AA310045) and the Association Foundation Program of Yunnan Provincial Science and Technology Department and Kunming Medical University (202001AY070001-282, 202301AY070001-006).

Acknowledgments

We should like to thank the cardiovascular surgery team collaborating to complete the surgery.

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

References

1. Hirst AE Jr., Johns VJ Jr., Kime SW Jr. Dissecting aneurysm of the aorta: a review of 505 cases. *Medicine (Baltimore)*. (1958) 37(3):217–79. doi: 10.1097/00005792-195809000-00003

2. Harris KM, Nienaber CA, Peterson MD, Woznicki EM, Braverman AC, Trimarchi S, et al. Early mortality in type a acute aortic dissection: insights from the international registry of acute aortic dissection. *JAMA Cardiol*. (2022) 7(10):1009–15. doi: 10.1001/jamacardio.2022.2718

3. Norton EL, Khaja MS, Williams DM, Yang B. Type a aortic dissection complicated by malperfusion syndrome. *Curr Opin Cardiol*. (2019) 34(6):610–5. doi: 10.1097/HCO.0000000000000667

4. Bayamin K, Power A, Chu MWA, Dubois L, Valdis M. Malperfusion syndrome in acute type a aortic dissection: thinking beyond the proximal repair. *J Card Surg*. (2022) 37(11):3827–34. doi: 10.1111/jocs.16872

5. Williams IM, Idhrees M, Velayudhan B, Bailey DM, Bashir M. Malperfusion syndrome in acute type a aortic dissection: it's not what you know, it's what you can prove!. *J Card Surg*. (2022) 37(11):3835–7. doi: 10.1111/jocs.16869

6. Geirsson A, Szeto WY, Pochettino A, McGarvey ML, Keane MG, Woo YJ, et al. Significance of malperfusion syndromes prior to contemporary surgical repair for acute type a dissection: outcomes and need for additional revascularizations. *Eur J Cardiothorac Surg*. (2007) 32(2):255–62. doi: 10.1016/j.ejcts.2007.04.012

7. Deeb GM, Williams DM, Bolling SF, Quint LE, Monaghan H, Sievers J, et al. Surgical delay for acute type a dissection with malperfusion. *Ann Thorac Surg*. (1997) 64(6):1669–75. doi: 10.1016/S0003-4975(97)01100-4

8. Berretta P, Trimarchi S, Patel HJ, Gleason TG, Eagle KA, Di Eusanio M. Malperfusion syndromes in type a aortic dissection: what we have learned from irad. *J Vis Surg*. (2018) 4:65. doi: 10.21037/jovs.2018.03.13

9. Chen S, Peng H, Zhuang H, Wang J, Yan P, Zhang W, et al. Optimized strategy to improve the outcomes of acute type a aortic dissection with malperfusion syndrome. *J Thorac Cardiovasc Surg*. (2024):S0022-5223(24)00019-9. doi: 10.1016/j.jtcvs.2024.01.006

10. Benbouchta K, Berrajaa M, Ofkire M, Ouafi NE, Bazid Z. An uncommon acute type a aortic dissection mimicking an inferior stemi. *Pan Afr Med J*. (2020) 36:247. doi: 10.11604/pamj.2020.36.247.23821

11. Wang Y, Zhu Z, Xu R, Li D, Wang T, Liu K. A complete occlusion of right coronary artery due to Stanford type a aortic dissection - successful treatment with extracorporeal membrane oxygenation (ecmo). *Braz J Cardiovasc Surg*. (2019) 34(4):491–4. doi: 10.21470/1678-9741-2018-0060

12. Yu J, Yu W, Zeng H, Gao J, Xiong J. Case report: surgery combined with extracorporeal membrane oxygenation for a patient with type a aortic dissection complicated with myocardial infarction after percutaneous coronary intervention. *Front Cardiovasc Med*. (2023) 10:1205373. doi: 10.3389/fcvm.2023.1205373

13. Coselli JS. Treatment of acute aortic dissection involving the right coronary artery and aortic valve. *J Cardiovasc Surg (Torino)*. (1990) 31(3):305–9.

14. Kawahito K, Adachi H, Murata S, Yamaguchi A, Ino T. Coronary malperfusion due to type a aortic dissection: mechanism and surgical management. *Ann Thorac Surg*. (2003) 76(5):1471–6. doi: 10.1016/S0003-4975(03)00899-3

15. Uchida K, Karube N, Minami T, Cho T, Matsuki Y, Nemoto H, et al. Treatment of coronary malperfusion in type a acute aortic dissection. *Gen Thorac Cardiovasc Surg*. (2018) 66(11):621–5. doi: 10.1007/s11748-018-1014-y

16. Taguchi Y, Kubo S, Ikuta A, Osakada K, Takamatsu M, Takahashi K, et al. Percutaneous coronary intervention for left main coronary artery malperfusion in acute type a aortic dissection. *Cardiovasc Interv Ther*. (2022) 37(2):333–42. doi: 10.1007/s12928-021-00793-4

17. Moreno PR, Fuster V. Jacc focus seminar on mechanical complications of acute myocardial infarction. *J Am Coll Cardiol*. (2024) 83(18):1775–8. doi: 10.1016/j.jacc.2024.03.414

18. Neri E, Toscano T, Papalia U, Frati G, Massetti M, Capannini G, et al. Proximal aortic dissection with coronary malperfusion: presentation, management, and outcome. *J Thorac Cardiovasc Surg*. (2001) 121(3):552–60. doi: 10.1067/mtc.2001.112534

19. Tong G, Wu J, Chen Z, Zhuang D, Zhao S, Liu Y, et al. Coronary malperfusion secondary to acute type a aortic dissection: surgical management based on a modified neri classification. *J Clin Med*. (2022) 11(6). doi: 10.3390/jcm11061693

20. Kreibich M, Bavaria JE, Branchetti E, Brown CR, Chen Z, Khurshan F, et al. Management of patients with coronary artery malperfusion secondary to type a aortic dissection. *Ann Thorac Surg*. (2019) 107(4):1174–80. doi: 10.1016/j.athoracsur.2018.09.065

21. Yau TH, Chong MH, Brigden ZM, Ngemoh D, Harky A, Bin Saeid J. The timing of surgical revascularization in acute myocardial infarction: when should we intervene? *J Cardiovasc Surg (Torino)*. (2022) 63(2):179–86. doi: 10.23736/s0021-9509.21.11984-6

22. Thiele H, Zeymer U, Neumann FJ, Ferenc M, Olbrich HG, Hausleiter J, et al. Intra-Aortic balloon counterpulsation in acute myocardial infarction complicated by cardiogenic shock (iabp-shock ii): final 12 month results of a randomised, open-label trial. *Lancet*. (2013) 382(9905):1638–45. doi: 10.1016/S0140-6736(13)61783-3

23. Ibanez B, James S, Agewall S, Antunes MJ, Bucciarelli-Ducci C, Bueno H, et al. 2017 Esc guidelines for the management of acute myocardial infarction in patients presenting with st-segment elevation: the task force for the management of acute myocardial infarction in patients presenting with st-segment elevation of the European Society of Cardiology (esc). *Eur Heart J*. (2018) 39(2):119–77. doi: 10.1093/eurheartj/ehx393

frontiers | Frontiers in Cardiovascular Medicine

TYPE Case Report
PUBLISHED 06 February 2025
DOI 10.3389/fcvm.2025.1516152

OPEN ACCESS

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RECEIVED 23 October 2024
ACCEPTED 15 January 2025
PUBLISHED 06 February 2025

CITATION
Bidovec J, Risteski P, Stähli BE, Dzemali O, Hofmann M and Stehli J (2025) Case Report: PCI of the left coronary artery after salvage operation in a comatose patient with an acute type A aortic dissection.
Front. Cardiovasc. Med. 12:1516152.
doi: 10.3389/fcvm.2025.1516152

Case Report: PCI of the left coronary artery after salvage operation in a comatose patient with an acute type A aortic dissection

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A 74-year-old female was found hemiplegic in a public restroom. After arrival in our stroke unit, a computed tomography (CT) was performed, and she was diagnosed with bilateral carotid artery and right vertebral artery occlusion due to an acute type A aortic dissection. The patient deteriorated quickly to a GCS of 3 and was brought to the operating room, where a salvage replacement of the ascending aorta and the proximal aortic arch was performed with unilateral antegrade cerebral perfusion. Three days later, a significant decrease in left ventricular function and increase in cardiac biomarkers were observed. Coronary CT displayed residual dissection of the aortic root, extending into the left main coronary artery. The patient underwent an intravascular ultrasound-guided stenting of the left main, resulting in total recovery of heart function. She was extubated on the fourth postoperative day, with no residual neurological impairment. This case report advocates for the proper management of patients with ATAAD with severe neurological impairment, emphasizing the importance of a robust multidisciplinary approach in their care.

KEYWORDS

aortic dissection, cerebral malperfusion, carotid obstruction, left main stenting, coronary artery dissection

1 Introduction

Preoperative cerebral malperfusion in patients with an acute type A aortic dissection (ATAAD) is associated with poor postoperative outcomes. The prognosis of comatose patients with bilateral carotid artery obstruction and last-remaining-vessel cerebral perfusion is particularly desolate (1–3). The International Registry of Acute Aortic Dissection (IRAD) investigators report a perioperative mortality over 60% in comatose

Abbreviations
ATAAD, acute type A aortic dissection; CT, computed tomography; GCS, Glasgow Coma Scale; ICU, intensive care unit; LAD, left anterior descending artery; LCX, left circumflex artery; NIRS, near infrared spectroscopy.

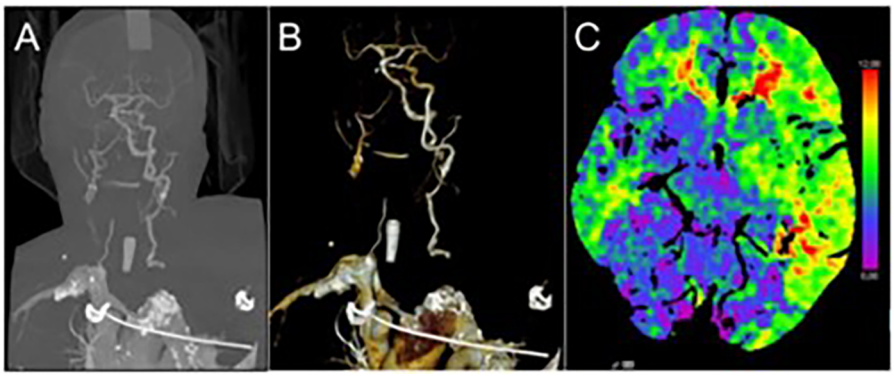

FIGURE 1
Preoperative contrast-enhanced computed tomography of the supra-aortic rteries showing long-segment antegrade obstruction of both carotid arteries and the right vertebral artery (A,B) and cerebral perfusion deficit predominately over the right cerebral hemisphere (C).

patients undergoing surgery for ATAAD (2). As a result, many patients are often deemed inoperable due to indicated poor postoperative outcomes. This case stands out due to the successful surgical procedure in comatose patient, which includes postoperative obstruction of the left coronary ostium and successful ultrasound-guided intervention in the left coronary artery.

2 Case description

A 74-year-old female with a history of arterial hypertension and previous stroke without residual deficits, was admitted to the emergency department. She was found in a public restroom, disoriented, aphasic and left-sided hemiplegic. The initial GCS was 13. After transfer to our stroke unit, a CT scan showed an ATAAD with a large primary entry in the ascending aorta. In addition, long-segment obstructions of both carotid arteries and the right vertebral artery were found, with a remaining cerebral perfusion through the left vertebral artery as the final remaining vessel (Figures 1A,B). This resulted in a pronounced perfusion deficit in the right cerebral hemisphere, with a mismatch of 355 ml (Figure 1C). After arrival, she deteriorated rapidly to a GCS of 3 and was taken straight to the operating theater, which was approximately 3.5 h after symptom onset. The estimated EuroSCORE II value was 58%.

3 Timeline

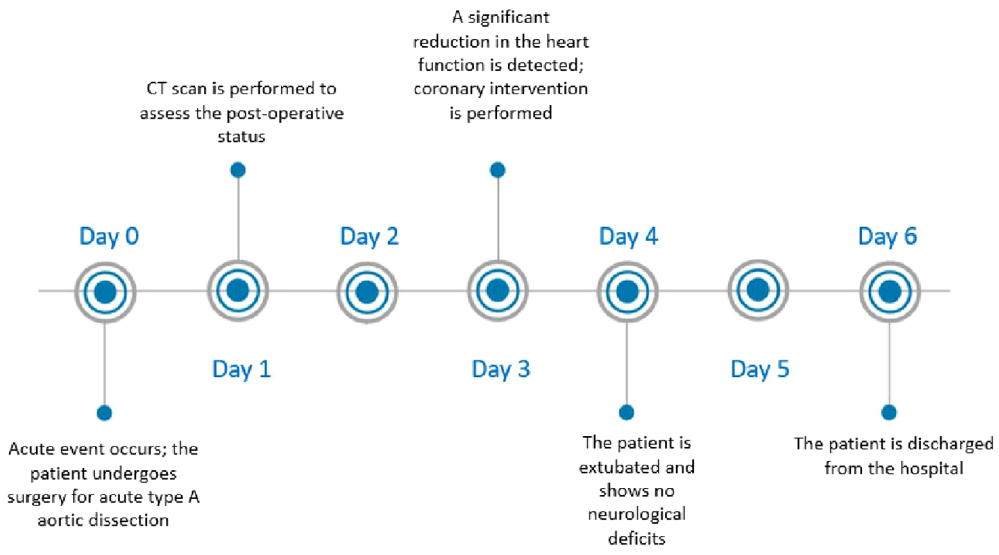

FIGURE 2
Case report timeline description.

4 Diagnostic assessment and treatment

After rapid-sequence intubation and instillation of a central venous and arterial lines, we cannulated the right axillary artery with an 18 French flexible cannula since this artery was not dissected. Following median sternotomy and pericardiotomy, the right atrial appendage was cannulated with a double-stage venous cannula, and cardiopulmonary bypass was initiated. We aimed for uninterrupted cerebral perfusion for the duration of the operation with antegrade flow via the right axillary artery and the circle of Wills as a primary collateral pathway. The adequacy of the cerebral perfusion (Figure 3) was confirmed by observing an immediate improvement of the Near Infrared Spectroscopy (NIRS) readings over both frontal cortices, which remained stable throughout the surgery.

Systemic cooling was limited to 26°C core temperature to preserve the cerebral autoregulation. The cerebral perfusion pressure was kept between 60 and 70 mmHg, as measured using the left radial arterial catheter. Using the pH-Stat method, moderate hypercapnia was permitted to decrease cerebral vascular resistance and improve cerebral perfusion. Intraoperatively, after clamping the ascending aorta, retrograde cardioplegia was initiated using a 14 French balloon-tipped flexible cannula. Then, the tubular aorta was divided.

The commissures of the aortic valve were exposed and reinforced with pledgeted commissural sutures. No glue or other sealants were used.

After inspecting the coronary ostia, cardioplegia was administered in a selective antegrade fashion using silicon-tipped cannulas. Upon achieving diastolic cardiac arrest, attention was focused on the aortic root, as a target body temperature of 26 °C had not yet been reached. During this time, the tubular part of the ascending aorta was removed, and the aortic root was closely inspected, which in our opinion did not require replacement. Once the target core temperature was achieved, the silastic bands were snared, and unilateral brain perfusion was maintained through the axillary artery in an antegrade fashion via the right vertebral and carotid arteries, without a decrease in NIRS measurements.

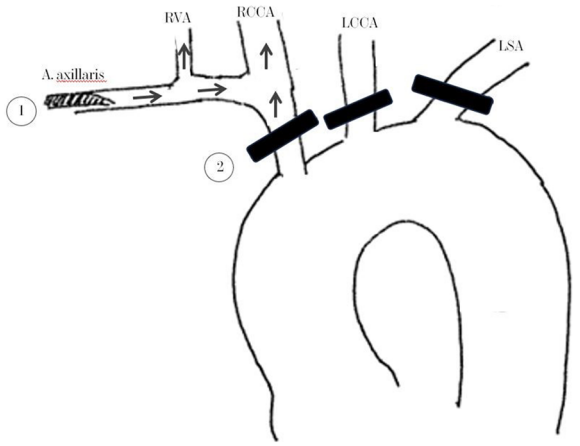

FIGURE 3
Schematic illustration of the arterial cannulation and the cerebral perfusion technique. (1) Positioning of the 18 French Cannula, (2) Black stripes show the positioning of the double encircled silastic bands "Vessel-loop" for vascular occlusion. Arrows show the direction of blood flow. RVA, right vertebral artery; RCCA, right common carotid artery; LCCA, left common carotid artery; LSA, left subclavian artery.

Hemiarch replacement with a polyester prosthesis was performed with a 5/0 prolene suture within 9 min of antegrade cerebral perfusion. We documented 32 min of myocardial ischemia and 87 min of cardiopulmonary bypass (CPB). Following the aortic repair, doppler ultrasound of the carotid arteries showed normal antegrade perfusion bilaterally with an adequate NIRS reading. Rewarming, weaning from the heart lung machine, and meticulous hemostasis was achieved.

Postoperatively, the patient was transferred to our intensive care unit (ICU), intubated and remained stable under low-dose vasopressors. The first ECG showed ST segment depressions in I, aVL, and V3–V6, which normalized the following day. The cardiac biomarkers demonstrated a decline in the slope early postoperatively. Partial respiratory insufficiency delayed extubation.

A CT angiography showed reestablished normal perfusion of both carotid arteries and a localized dissection of the aortic root with perpetuation into the left main coronary artery (Figure 4). Due to the declining levels of cardiac biomarkers, an unknown neurological outcome, and improvements in hemodynamic stability, no further measures were implemented on the first postoperative day. However, on the third postoperative day new T-wave inversions in leads V3–V6, accompanied by a progressive increase in cardiac biomarkers, were noticed. A transthoracic echocardiographic exam demonstrated a decline in left ventricular ejection fraction from 60% to 25% with global hypokinesia and new moderate mitral valve regurgitation. Because of the rapid hemodynamic deterioration, the only logical explanation for a sudden drop in ejection fracture due to luminal narrowing of the left main ostium. It was presumed that this was most likely because of dynamic changes of the residual dissection in the aortic root or through a hematoma compressing from the outside. Following an interdisciplinary discussion, we decided to proceed with an invasive coronary angiogram, despite the known risks associated with percutaneous coronary intervention (PCI) in cases of coronary dissection, for the following reasons: First, the patient exhibited clear signs of myocardial ischemia. Second, the 90% stenosis of the left main artery posed a significant risk of complete occlusion at any moment, which could result in sudden cardiac death for the patient.

After gaining radial access, a 6 French Judkins left 4.0 guide was advanced to the left main ostium. To prevent the dissection progression, both ostia left anterior descending artery (LAD) and the left circumflex artery (LCX) had to be secured with a stent. The guidewire was advanced in the mentioned arteries without any contrast injection. Correct position of the wires in the true lumen was confirmed by intravascular ultrasound (Figure 5). After wiring the LAD and the LCX, the first contrast injection confirmed the dissection with contrast dye spreading in the false lumen and retrograde towards the aortic root. With no further delay, LAD, and LCX were stented using a nano-crush technique to the edge of the LM ostium without protrusion into the aorta.

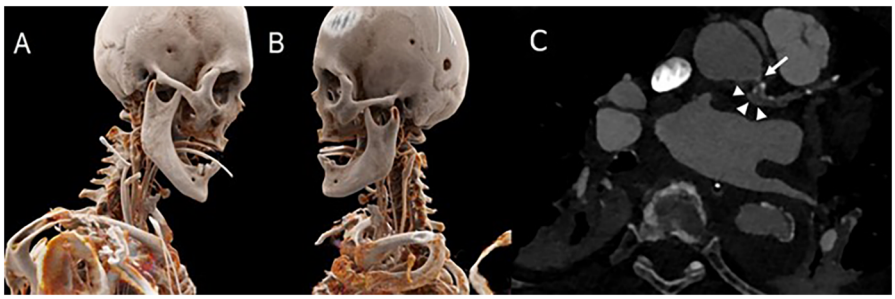

FIGURE 4
Postoperative CT-scan showing 3-D reconstructions of patient right **(A)** and left **(B)** A CT angiogram displaying a dissection membrane narrowing of the left main ostium (white arrow) and the false lumen (white arrowheads) **(C)**.

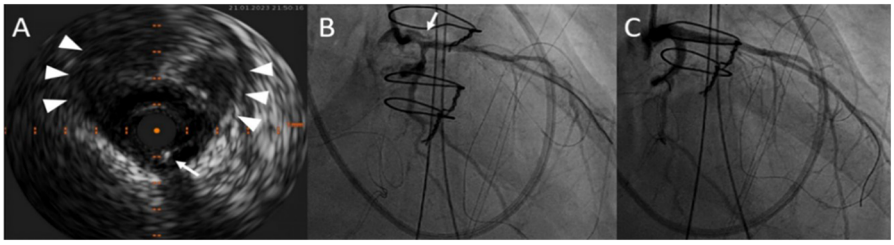

FIGURE 5
After wiring both the left anterior descending artery and the left circumflex artery, intravascular ultrasound of the left main coronary artery confirmed the correct wire position (white arrows) in the true lumen. The white arrowheads point at the large false lumen **(A)**. RAO projection in the coronary angiogram displaying dissection narrowing of the left main ostium and white arrow points at the false lumen **(B)**. Postoperative result after LAD and LCX stenting with a nano-crush technique **(C)**.

A Boston Scientific Synergy Megatron 3.5/20 mm stent was used for the LAD, and a Boston Scientific Synergy 3.0/16 mm for the LCX with excellent results. To better assess the result, we performed the IVUS again after stent deployment. The patient was given an antiplatelet therapy with 500 mg Aspirin IV at the start of the procedure and a loading dose of 600 mg Clopidogrel after the PCI. After the intervention, an improvement of the ejection fraction to 40% and a reduction of the residual mitral regurgitation from severe to mild was noticed. The patient was extubated on the fourth postoperative day without neurological impairment. She was transferred to the rehabilitation center on the sixth postoperative day (Figure 2) and returned home on the 42nd postoperative day after she made a full recovery. The patient was maintained on dual antiplatelet therapy consisting of Aspirin and Clopidogrel for 12 months.

5 Discussion

ATAAD with a global neurologic deficit is associated with very high mortality and morbidity (1–3). Comatose patients with single-vessel cerebral perfusion very rarely survive the acute event. They are often declared inoperable (4), even though some studies suggest that an early intervention may lead to a postoperative improvement of the neurologic deficit (5–9). Further, the results of the IRAD suggests that an ongoing brain ischemia should not be a contraindication for the surgery, especially if the patient does not display definitive signs of severe irreversible neurological damage (2). The decision to operate our patient was taken after observing short periods of cognitive bursts and alternating neurologic symptoms, which most likely was due to a dynamic low-flow and not a complete interruption of the perfusion to the brain.

It is our belief that cannulating the right axillary artery could be the most expedient method to restore antegrade cerebral perfusion, especially in cases where the dissection membrane occlude the ostia of supra-aortic branches. The real-time NIRS findings supported our judgment, as the regional oxygen saturation improved once the patient was placed on the extracorporeal circulation using axillary cannulation. Additionally, intraoperative carotid Doppler sonography confirmed sustained extracranial perfusion. Nevertheless, it is critical not to overlook the aortic root and coronary ostia after reestablishing brain perfusion, given that coronary involvement in ATAAD significantly predicts both early and late mortality (10, 11).

However, when neurologic symptoms are present without evident myocardial compromise, priority shifts towards re-establishing cerebral perfusion. Unfortunately, the preoperative CT scan did not include the aortic root because of the neurologist's initial focus. During the operation, we did not observe any signs of aortic root dissection, which is why we did not underwent aortic root replacement. In our opinion, the root dissection likely occurred postoperatively, and was subsequently detected in the postoperative

CT scan. The weaning of heart lung machine was uneventful, and we saw no significant echocardiographic or ECG changes that would indicate myocardial ischemia due to the left main obstruction. In retrospect, an earlier intervention upon detecting the narrowing of the left main artery would have prevented the subsequent hemodynamic deterioration. However, our approach was aimed at simplifying the post-operative course. The argument stands that intraoperative coronary angiography could offer valuable insights into myocardial perfusion, potentially improving postoperative outcomes without adding procedural complexity (12).

Upon detecting these signs of myocardial ischemia, PCI for the left main dissection was undertaken despite the inherent risks associated with treating coronary dissections. In conclusion, this case underscores the effectiveness of a multidisciplinary approach and feasibility of PCI for left main dissection in a challenging cardiovascular emergency.

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

JB: Conceptualization, Methodology, Project administration, Writing – original draft, Writing – review & editing. PR: Supervision, Writing – review & editing. BS: Supervision, Writing – review & editing. OD: Supervision, Validation, Writing – review & editing. MH: Writing – review & editing. JS: Supervision, Validation, Writing – review & editing.

Funding

The authors declare that no financial support has been received for the research, authorship, and/or publication of this article.

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.

The author(s) declared that they were an editorial board member of Frontiers, at the time of submission. This had no impact on the peer review process and the final decision.

Generative AI statement

The author(s) declare that no Generative AI was used in the creation of this manuscript.

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References

1. Kreibich M, Desai ND, Bavaria JE, Szeto WY, Vallabhajosyula P, Itagaki R, et al. Preoperative neurological deficit in acute type A aortic dissection. *Interact Cardiovasc Thorac Surg.* (2020) 30(4):613–9. doi: 10.1093/icvts/ivz311

2. Di Eusanio M, Patel HJ, Nienaber CA, Montgomery DM, Korach A, Sundt TM, et al. Patients with type A acute aortic dissection presenting with major brain injury: should we operate on them? *J Thorac Cardiovasc Surg.* (2013) 145(3):S213–21. doi: 10.1016/j.jtcvs.2012.11.054

3. Aita JF, Agripnidis T, Testud B, Barral PA, Jacquier A, Reyre A, et al. Stenting in brain hemodynamic injury of carotid origin caused by type A aortic dissection: local experience and systematic literature review. *J Pers Med.* (2022) 13(1):58. doi: 10.3390/jpm13010058

4. Tanaka H, Okada K, Yamashita T, Morimoto Y, Kawanishi Y, Okita Y. Surgical results of acute aortic dissection complicated with cerebral malperfusion. *Ann Thorac Surg.* (2005) 80:726. doi: 10.1016/j.athoracsur.2004.01.038

5. Fukuhara S, Norton EL, Chaudhary N, Burris N, Shiomi S, Kim KM, et al. Type A aortic dissection with cerebral malperfusion: new insights. *Ann Thorac Surg.* (2021) 112(2):501–9. doi: 10.1016/j.athoracsur.2020.08.046

6. Tsukube T, Hayashi T, Kawahira T, Haraguchi T, Matsukawa R, Kozawa S, et al. Neurological outcomes after immediate aortic repair for acute type A aortic dissection complicated by coma. *Circulation.* (2011) 124(11_suppl_1):S163–7. doi: 10.1161/CIRCULATIONAHA.110.011551

7. Pocar M, Passolunghi D, Moneta A, Donatelli F. Recovery of severe neurological dysfunction after restoration of cerebral blood flow in acute aortic dissection. *Interact Cardiovasc Thorac Surg.* (2010) 10:839–41. doi: 10.1510/icvts.2009.228908

8. Pocar M, Passolunghi D, Moneta A, Mattioli R, Donatelli F. Coma might not preclude emergency operation in acute aortic dissection. *Ann Thorac Surg.* (2006) 81(4):1348–51. doi: 10.1016/j.athoracsur.2005.09.076

9. Sasaki H, Harada T, Ishitoya H, Sasaki O. Aorto-carotid bypass for type A acute aortic dissection complicated with carotid artery occlusion: no touch until circulatory arrest. *Interact Cardiovasc Thorac Surg.* (2020) 31:263–5. doi: 10.1093/icvts/ivaa092

10. Rizzo RJ, Aranki SF, Aklog L, Couper GS, Adams DH, Collins JJ Jr, et al. Rapid noninvasive diagnosis and surgical repair of acute ascending aortic dissection. Improved survival with less angiography. *J Thorac Cardiovasc Surg.* (1994) 108(3):567–75. doi: 10.1016/S0022-5223(94)70269-1

11. Kern MJ, Serota H, Callicoat P, Deligonul U, Lee WH, Aguirre F, et al. Use of coronary arteriography in the preoperative management of patients undergoing urgent repair of the thoracic aorta. *Am Heart.* (1990) 119(1):143–8. doi: 10.1016/S0002-8703(05)80094-8

12. Yang J, Li X, Chen Z, Sun T, Fan R, Yu C. Case report: intraoperative open-heart coronary angiography in acute type A aortic dissection. *Front Cardiovasc Med.* (2021) 8:731581. doi: 10.3389/fcvm.2021.731581

frontiers | Frontiers in Cardiovascular Medicine

TYPE Case Report
PUBLISHED 07 February 2025
DOI 10.3389/fcvm.2025.1486771

OPEN ACCESS

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RECEIVED 26 August 2024
ACCEPTED 20 January 2025
PUBLISHED 07 February 2025

CITATION
Çetinarslan Ö and Saba D (2025) Case Report: Staged surgical management in ESRD: off-pump CABG followed by renal transplantation to enhance graft survival. Front. Cardiovasc. Med. 12:1486771. doi: 10.3389/fcvm.2025.1486771

Case Report: Staged surgical management in ESRD: off-pump CABG followed by renal transplantation to enhance graft survival

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Patients with end-stage renal disease face a significantly higher risk of cardiovascular diseases. For patients who are candidates for renal transplantation (RT), major surgeries such as coronary artery bypass grafting (CABG) are associated with cardiac complications as well as higher rates of post-operative complications, including the need for large amounts of blood transfusion, worsening kidney function, infection, and graft rejection. Studies have shown that blood transfusions can increase the risk of graft rejection due to immune system activation. Off-pump CABG (OPCAB), also known as beating heart surgery, is a technique in which a heart–lung machine is not used, and the heart continues to beat throughout the procedure. The main advantage of OPCAB surgery compared to on-pump CABG (ONCAB) is that it requires fewer blood product transfusions and has fewer renal, pulmonary, and hematological complications. This case series uniquely discusses two patients who underwent successful beating heart CABG without blood transfusion, followed by RT.

KEYWORDS

end-stage renal disease (ESRD), off-pump coronary artery bypass grafting (OPCAB), on-pump coronary artery bypass grafting (ONCAB), renal transplantation (RT), staged surgery vs. combined surgery

Introduction

Patients with end-stage renal disease (ESRD) face a significantly higher risk of cardiovascular diseases (1). In this population, coronary artery disease (CAD) is driven by complex mechanisms. Excessive medial calcinosis, characterized by calcium deposition in the vascular smooth muscle layer, due to chronic inflammation and imbalances in calcium-phosphate metabolism leads to arterial stiffness and impaired vascular compliance, significantly increasing cardiovascular endpoints in this population. The presence of CAD in patients with ESRD often necessitates coronary artery bypass grafting (CABG) prior to renal transplantation (RT). Unfortunately, in patients who are candidates for RT, major surgeries such as CABG are associated with cardiac complications, worsening kidney function, post-operative infections, and graft rejection. Studies have shown that blood transfusions not only complicate post-operative recovery but also increase the risk of graft rejection due to immune system activation (2, 3).

Off-pump CABG (OPCAB), also known as beating heart surgery, is a technique in which the heart–lung machine is not used, and the heart continues to beat throughout the procedure. The main advantage of OPCAB surgery compared to on-pump CABG (ONCAB) is that it requires fewer blood product transfusions and has fewer renal, pulmonary, and hematological complications (4).

This case series uniquely discusses two patients who underwent successful OPCAB without blood transfusion, followed by RT. This case series also aims to provide a perspective on the debate between combined and staged CABG and RT, despite the increasing popularity of combined surgeries. Written informed consent was obtained from all patients.

Case 1

A 55-year-old male patient was referred to our transplantation clinic. The patient was on a 3/7 dialysis program. During his cardiac assessment, electrocardiogram (ECG) findings were non-significant, and his left ventricular ejection fraction (LVEF) was 58%. He was on colchicine treatment for mild pleural and pericardial effusion. Myocardial perfusion scintigraphy (MPS) revealed anterior and apical non-reversible and reversible perfusion defects. Three-vessel CAD was diagnosed by coronary angiography (CAG) (Figures 1, 2). An off-pump CABGx3 [the left internal mammary artery to the left anterior descending artery (LIMA-LAD), aort to right posterior descending artery (Ao-RPD), Ao to second optuse marginal branch of circumflex artery (Ao-CxOM2)] was performed without the use of blood products. The patient's pre-CABG laboratory values were as follows: hemoglobin (Hgb) 8.5 g/dl, hematocrit (Hct) 24.6%, platelets (PLT) 172,000/µl, and creatinine (Cr) 6.67 mg/dl. Post-CABG, the following values were measured: Hgb 7.61 g/dl, Hct 22.8%, PLT 146,000/µl, and Cr 9 mg/dl. On post-CABG day 5, the patient was transferred to nephrology while continuing oral cardiac treatment.

The patient was subsequently hospitalized at an external center due to decompensation and pancytopenia. During the etiological investigation, potential causes such as marrow suppression, nutritional deficiencies, and drug reactions were explored. Colchicine-related pancytopenia was also considered. After discontinuation of colchicine, the pancytopenia resolved. After a 1-month hospital stay involving intensive hemodialysis and close hematological follow-up, preparations for RT began. Serological tests were performed, and panel reactive antibodies (PRA) were in normal range. Successful RT from a living donor was performed on the 60th day post-CABG. The immunosuppressive and prophylactic treatment protocols in our RT clinic are as follows: intravenous prednisolone administration started with 1,000 mg on the preoperative day, 500 mg intra-operatively, 250 mg on post-operative day (POD) 1, 125 mg on POD 2, 100 mg on POD 3, 80 mg on POD 4, 60 mg on POD 5, and 40 mg on POD 6. Following discharge, oral prednisolone was prescribed at 20 mg daily for the first month, 15 mg daily for the second month, 10 mg daily for the third month, and then maintained lifelong at 5 mg daily.

In addition to steroid therapy, doses of tacrolimus and mycophenolate sodium were adjusted based on tacrolimus level monitoring. Due to the recurrence of neutropenia in the third month following RT, mycophenolate sodium was discontinued, and everolimus therapy was initiated, with dosing guided by serum level monitoring.

At the 12-month follow-up, the patient was found to be both cardiac and renal stable.

Case 2

A 58-year-old male with hypertension and secondary diabetes due to previous steroid treatment in his medical history was referred for cardiac evaluation while preparing for RT. He had been diagnosed with focal segmental glomerulosclerosis (FSGS)

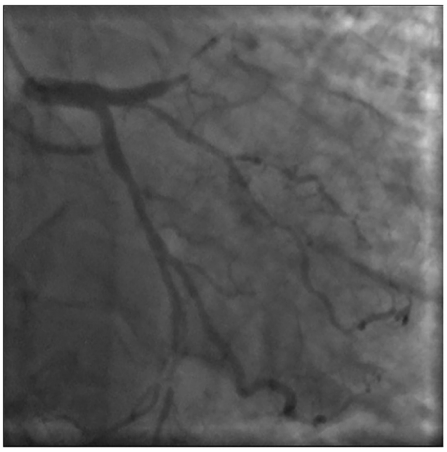

FIGURE 1
Subtotal occlusion of the LAD and Cx-OM2 artery in a coronary angiogram.

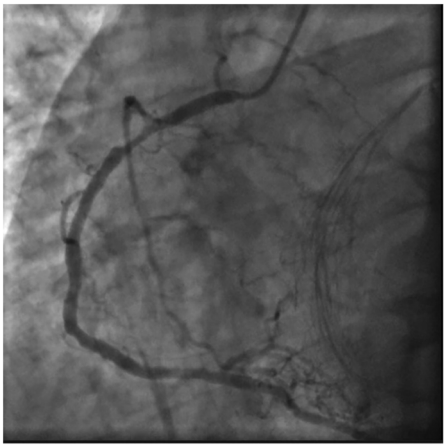

FIGURE 2
Coronary angiography view of RCA proximal 90% stenosis.

3 years prior and treated with steroids for 5 months. Transthoracic echocardiography (TTE) and ECG findings were non-significant. CAG revealed three-vessel CAD. An off-pump CABGx3 [the right internal mammary artery to LAD (RIMA-LAD), LIMA to first diagonal and optuse marginal branch (LIMAD1-OM in a saphenous Y graft configuration)] was performed because of the porcelain aorta without blood transfusion. Pre-CABG laboratory values were as follows: Hgb 16.3 g/dl, Hct 47.6%, PLT 209,000/µl, and Cr 5.15 mg/dl. Post-CABG laboratory values were Hgb 12.5 g/dl, Hct 38%, PLT 166,000/µl, and Cr 6.33. Perioperative routine prophylaxis with valganciclovir 450 mg 1 × 1 p.o. and sulfamethoxazole/trimethoprim 80/400 mg 1 × 1 p.o. was applied.

There was no significant difference in post-CABG TTE. On post-CABG day 5, the patient was discharged with the following treatment regimen: acetylsalicylic acid 100 mg once daily, metoprolol 50 mg once daily, and lercanidipine 10 mg once daily.

The patient was placed on the 2/7 hemodialysis program after CABG due to worsening kidney function. Creatinine clearance, 24-h proteinuria, ultrasound imaging of the kidneys, and serological tests were re-evaluated. PRA levels were within the normal range. Successful RT from a living donor was performed on the 38th day post-CABG. Immediate graft function was achieved with adequate urine output post-surgery. Immunosuppressive treatment was arranged in accordance with the protocols of our transplant team. Three months of trimethoprim-sulfamethoxazole (TMP-SMX) 480 mg daily against *Pneumocystis jirovecii* and valganciclovir 450 mg daily against cytomegalovirus (CMV) were prescribed as prophylaxis.

At 12-month follow-up, the patient was found to be both cardiac and renal stable.

Discussion

Patients with ESRD have a higher risk of cardiovascular disease compared to the general population, and cardiac surgery carries a higher risk. Moreover, CAD and heart surgery can negatively affect the early and late outcomes of RT.[1]

CABG remains a cornerstone in the management of multi-vessel CAD (5). It offers superior revascularization in cases of multi-vessel CAD due to its ability to bypass complex and diffusely calcified lesions, which are common in ESRD and RT populations. Meta-analyses have consistently shown that CABG reduces the risk of major adverse cardiac and cerebrovascular events (MACCEs), including myocardial infarction and repeat revascularization, compared to PCI. For our young RT recipients, this durability is especially critical given their longer expected lifespan compared to dialysis-dependent patients (6–8). Advances in surgical techniques have resulted in two principal approaches: OPCAB, which is performed on a beating heart without the use of cardiopulmonary bypass (CPB), and ONCAB, which employs CPB and cardioplegic arrest. Each method has distinct advantages and limitations, often tailored to the patient's comorbidities and anatomy and surgeon's expertise.

OPCAB procedures, similar to ONCAB, were performed under general anesthesia via median sternotomy. Stabilization of the target vessels was accomplished with the Acrobat-i stabilizer (Getinge). Hemodynamic stability during heart positioning for distal anastomosis was maintained using the Trendelenburg position and pharmacologic management with intravenous noradrenaline infusion as needed. Anticoagulation was achieved by administering intravenous heparin at 1.5 mg/kg of body weight, ensuring an activated clotting time (ACT) above 300 s throughout the procedure. To improve surgical field visibility, proximal occlusion of the target vessel was performed with a micro-vessel occluder, and a sterile medical air blower was used to clear blood from the area. The distal artery was routinely kept open, and neither ischemic preconditioning nor intracoronary shunts were used. Whenever possible, the LAD was grafted first to restore blood flow to the anterior wall as early as possible (9). Anastomoses to the right coronary artery (RCA) were avoided unless critical stenosis (>90%) was present. Unless there is a specific indication, we do not perform thoracic CT prior to CAB surgery. Of course, epiaortic ultrasound evaluation of the ascending aorta is more effective than palpation by the surgeon. However, in our second case, the porcelain aorta was so evident by palpation that, given our extensive experience with OPCAB, palpation alone was sufficient.

Multiple studies, including meta-analyses, indicate comparable 10-year survival rates between OPCAB and ONCAB. In addition, other long-term endpoints, including angina, graft patency, heart failure, rehospitalization, and stroke, suggest similar adverse events (10). A recent study involving 24,883 participants showed that the benefit of OPCAB on mortality at 8-year follow-up did not differ between diabetic and non-diabetic patients (11). On the other hand, another recent study found ONCAB superior to OFCAB due to shorter procedure time and fewer MACCEs (9, 12). These controversial results indicate that the operator's expertise plays a critical role, particularly for OPCAB, where surgeon skill significantly influences graft patency and overall outcomes.

OPCAB offers several advantages for ESRD patients awaiting RT. By avoiding CPB, OPCAB reduces the risk of systemic inflammation, coagulopathy, and the need for blood transfusions, thereby minimizing immunological challenges that could trigger graft rejection (13). Studies have highlighted the importance of minimizing blood transfusions to prevent immune system activation and potential graft rejection (14, 15). In addition, preoperative measurement of PRA levels is crucial for assessing the risk of graft rejection and tailoring immunosuppressive therapy (16). OPCAB also provides an advantage as a less invasive option in patients with severe aortic calcification, as in case 2.

The most recent propensity-match analysis comparing OPCAB and ONCAB in ESRD patients not on dialysis showed no

[1]Patients with end-stage renal disease have a higher risk of cardiovascular disease compared to the general population and cardiac surgery is very high risk. Moreover, cardiovascular diseases and heart surgery negatively affect the early and late results of kidney transplantation.

differences in mortality, stroke, and post-operative dialysis rates. On the other hand, longer hospital stays and re-operation due to bleeding/tamponade were reported more frequently in ONCAB (17). In patients with chronic kidney disease (CKD), even in the absence of RT, the frequent occurrence of comorbidities such as porcelain aorta (as seen in case 2), increased frailty, and polypharmacy make them more susceptible to drug side effects (as seen in case 1) and hematological complications. Consequently, OPCAB demonstrates significantly better outcomes than ONCAB in terms of short-term mortality (18).

In our practice, we prioritize the use of arterial grafts in high-risk patients with multiple comorbidities, aligning with recent studies demonstrating better clinical outcomes for patients with ESRD undergoing CABG when arterial grafts are used instead of saphenous vein grafts (19). For instance, the RAPCO trials reported a 29% reduction in MACCEs over 15 years when using radial artery grafts instead of saphenous vein grafts (20). However, in patients with ESRD, considering that dialysis and an AV fistula may still be required even after RT, we avoid using the radial approach in CAG and similarly refrain from using radial grafts in CABG.

The appropriate approach for patients with CAD who were scheduled for RT remains a topic of debate. Stabilizing coronary flow as a first step reduces the risk of myocardial ischemia during subsequent transplantation. This stabilization allows the heart to better cope with the increased hemodynamic burden of major surgery and intraoperative cardiac stress. For instance, pancytopenia following combined surgery, as seen in case 1, causes significant risks in terms of both graft patency and cardiac stability. Furthermore, it may necessitate undesirable modifications to immunosuppressive treatment regimens.

Managing two major surgeries simultaneously is associated with prolonged surgery duration and increased anesthesia exposure, raising challenges in the stabilization of blood pressure and intraoperative fluid balance, especially in ESRD patients. The interval between CABG and RT can optimize the patient's immunological status and reduce the risk of graft rejection. On the other hand, in the combined strategy, immunosuppressive therapy management may be more complex, potentially endangering both cardiac and renal functions. In addition, in this fragile patient group, as in the first case report, if a complication develops between two major surgeries, transplantation can be postponed until the patient is stable.

This case series is unique in presenting patients who underwent OPCAB followed by RT without blood transfusion, coupled with a 12-month follow-up period. By staging the procedures, we planned to reduce the risk of warm ischemia in the kidney, which may be caused by hypotension and prolonged surgery duration during a combined CABG and RT procedure. We present a different perspective suggesting that a staged approach may enable better management of complications in this patient group characterized by multiple comorbidities and polypharmacy. This approach may help preserve the viability of the graft, which is particularly valuable when obtained from a living donor.

A multidisciplinary team involving cardiology, cardiothoracic surgery, and transplant nephrology assessed both patients and determined that they were poor candidates for percutaneous coronary intervention (PCI) due to complex CAD, low hemoglobin levels, and a hypercoagulable state; consequently, decided on staged surgery. In the pre-RT cardiac evaluation of patients with ESRD, whose exercise capacity is already limited by many other accompanying chronic diseases, clinical suspicion by cardiology and its tests are diagnostic. The comprehensive approach provided by transplant nephrology ensures that patients have an uninterrupted follow-up process. Alongside an experienced clinical and surgical team, perhaps the most crucial point is to include the patient in this multidisciplinary decision-making process. We informed our patients comprehensively about both combined and staged surgery options and discussed the advantages and disadvantages of each approach.

Conclusion

Off-pump CABG followed by staged RT presents a beneficial approach for patients with ESRD and significant CAD. Common comorbidities in these patient groups, such as porcelain aorta (as seen in case 2), increased frailty, polypharmacy, and hematological complications, highlight the advantages of OFCAB. In the current era, where major surgeries performed in the same session are increasingly popular, this strategy provides a perspective that can prepare patients for a more successful RT and post-operative follow-up by ensuring immunological integrity and the complete wellbeing of patients. Each patient is unique, and the decision-making process should be individualized.

Limitations

This study has some significant limitations. Since it includes only two patients, the results cannot be applied to larger populations. In addition, the outcomes may be affected by the high level of experience of the transplant and surgical teams, as well as the specific facilities and resources available at our hospital, which may not be the same in other centers. Another limitation is that we did not routinely perform preoperative thoracic CT scans before CABG unless there was a clear indication. Although epiaortic ultrasound evaluation of the ascending aorta usually provides reliable information, in the second case, the porcelain aorta was so obvious by palpation that imaging was deemed unnecessary due to the team's significant OPCAB experience. However, in less experienced centers, imaging may be needed for better surgical planning. These points indicate that surgical strategies should be adapted based on the team's experience and hospital resources.

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

Ethical approval was not required for the study involving humans in accordance with the local legislation and institutional requirements. Written informed consent to participate in this study was not required from the participants or the participants' legal guardians/next of kin in accordance with the national legislation and the institutional requirements. 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

ÖÇ: Conceptualization, Data curation, Formal Analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing. DS: Investigation, Methodology, Project administration, Resources, Supervision, Writing – review & editing.

Funding

The author(s) declare that no financial support was received for the research, authorship, and/or publication of this article.

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. Bhatti NK, Galougahi KK, Paz Y, Nazif T, Moses JW, Leon MB, et al. Diagnosis and management of cardiovascular disease in advanced and end-stage renal disease. *J Am Heart Assoc*. (2016) 5(8):1–11. doi: 10.1161/JAHA.116.003648

2. Atılgan K, Demirdaş E, Boran M, Cengiz Z, Çiçekçioğlu F. Coronary artery bypass grafting in a renal transplant patient: case report. *Eur Res J*. (2019) 5(2):424–7. doi: 10.18621/eurj.386681

3. Jiang Y, Xu L, Liu Y, Deng B, Dong N, Chen S. Beating-heart on-pump coronary artery bypass grafting vs. off-pump coronary artery bypass grafting: a systematic review and meta-analysis. *J Thorac Dis*. (2021) 13(7):4185–94. doi: 10.21037/jtd-21-268

4. Al-Sabti H, Krishnan K. Beating heart surgery—current concept. *Oman Med J*. (2008) 23(4):233–5.

5. Omerovic E, Råmunddal T, Petursson P, Angerås O, Rawshani A, Jha S, et al. Percutaneous vs. surgical revascularization of non-ST-segment elevation myocardial infarction with multivessel disease: the SWEDEHEART registry. *Eur Heart J*. (2024) 27:ehae700. doi: 10.1093/eurheartj/ehae700

6. Tuttle ML, Weiner DE. Coronary artery bypass grafting versus percutaneous coronary intervention in patients receiving dialysis: is CABG worth the risk? *Kidney Med*. (2023) 6(2):100787. doi: 10.1016/j.xkme.2023.100787

7. Luo C, Wang Q, Nong S, et al. Meta-analysis of clinical adverse events after CABG vs. PCI in patients with chronic kidney disease and coronary artery disease. *BMC Cardiovasc Disord*. (2023) 23(590):4, 5. doi: 10.1186/s12872-023-03560-w

8. Bangalore S, Maron DJ, O'Brien SM, et al. Management of coronary disease in patients with advanced kidney disease. *N Engl J Med*. (2020) 382(17):1608–18. doi: 10.1056/NEJMoa1915925

9. Davit S, Senkaya I, Ercan AK, Kan II, Ozkan H. Is 100% beating heart coronary by-pass justified? *Cardiovasc Surg*. (2002) 10(6):579–85. Erratum in: *Cardiovasc Surg*. (2003) 11(1):102. doi: 10.1016/s0967-2109(02)00068-6

10. Chaudhry UA, Rao C, Harling L, Athanasiou T. Does off-pump coronary artery bypass graft surgery have a beneficial effect on long-term mortality and morbidity compared with on-pump coronary artery bypass graft surgery? *Interact Cardiovasc Thorac Surg*. (2014) 19(1):149–59. doi: 10.1093/icvts/ivu075

11. Adineh HA, Hoseini K, Zareban I, et al. Comparison of outcomes between off-pump and on-pump coronary artery bypass graft surgery using collaborative targeted maximum likelihood estimation. *Sci Rep*. (2024) 14:11373. doi: 10.1038/s41598-024-61846-1

12. Zubarevich A, Kadyraliev B, Arutyunyan V, Chragyan V, Askadinov M, Sozkov A, et al. On-pump versus off-pump coronary artery bypass surgery for multi-vessel coronary revascularization. *J Thorac Dis*. (2020) 12(10):5639–46. doi: 10.21037/jtd-20-1284

13. Atik FA, Borges CC, Ulhoa MB, Chaves RB, Barzilai VS, Biondi RS, et al. Combined heart and kidney transplantation: initial clinical experience. *Braz J Cardiovasc Surg*. (2022) 37(2):263–7. doi: 10.21470/1678-9741-2020-0720

14. Tekin S, Zengin M, Tekin İ, Yüceetin L, Yavuz HA, Okutan H, et al. Simultaneous cardiac surgery and renal transplantation compared with renal transplantation after cardiac surgery. *Transplant Proc*. (2015) 47(5):1340–4. doi: 10.1016/j.transproceed.2015.04.024

15. Awad MA, Czer LSC, Emerson D, Jordan S, De Robertis MA, Mirocha J, et al. Combined heart and kidney transplantation: clinical experience in 100 consecutive patients. *J Am Heart Assoc*. (2019) 8(4):e010570. doi: 10.1161/JAHA.118.010570

16. Gallo M, Trivedi JR, Schumer EM, Slaughter MS. Combined heart-kidney transplant versus sequential kidney transplant in heart transplant recipients. *J Card Fail*. (2020) 26(7):574–9. doi: 10.1016/j.cardfail.2020.03.002

17. Fudulu DP, Argyriou A, Kota R, Chan J, Vohra H, Caputo M, et al. Effect of on-pump vs. off-pump coronary artery bypass grafting in patients with non-dialysis-dependent severe renal impairment: propensity-matched analysis from the UK registry dataset. *Front Cardiovasc Med*. (2024) 11:1341123. doi: 10.3389/fcvm.2024.1341123

18. Ahmed M, Majeed K, Ali H, Syed H, Batool A. Off-pump vs. on-pump coronary artery bypass grafting in patients with chronic kidney disease: an updated systematic review and meta-analysis. *Int Urol Nephrol*. (2025) 57(2):463–77. doi: 10.1007/s11255-024-04198-z

19. Chandra R, Heid CA. The conduit conundrum: a commentary on "outcomes of radial artery versus saphenous vein as a second conduit after coronary artery bypass grafting". *Am J Cardiol*. (2024) 217:158–60. doi: 10.1016/j.amjcard.2024.02.018

20. Hamilton GW, Raman J, Moten S, Matalanis G, Rosalion A, on behalf of the RAPCO Investigators. Radial artery vs. internal thoracic artery or saphenous vein grafts: 15-year results of the RAPCO trials. *Eur Heart J*. (2023) 44(26):2406–8. doi: 10.1093/eurheartj/ehad108

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TYPE Case Report
PUBLISHED 13 February 2025
DOI 10.3389/fcvm.2025.1483395

OPEN ACCESS

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RECEIVED 30 September 2024
ACCEPTED 27 January 2025
PUBLISHED 13 February 2025

CITATION
Idu M, Lim CY and Chua KC (2025) Case Report: Late presentation of post-coronary artery bypass surgery pericardial effusion heralding mediastinitis with tracheocutaneous fistula.
Front. Cardiovasc. Med. 12:1483395.
doi: 10.3389/fcvm.2025.1483395

Case Report: Late presentation of post-coronary artery bypass surgery pericardial effusion heralding mediastinitis with tracheocutaneous fistula

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Post-coronary artery bypass surgery pericardial effusions are typically self-limiting but may rarely be significant, causing pericardial tamponade. We describe a case of late post-operative pericardial effusion that required pericardiocentesis and subsequent readmission for mediastinitis with the added complication of a tracheocutaneous fistula. Our case report is the first reported instance of pericardial tamponade heralding the onset of mediastinitis. It also describes the rare complication of tracheocutaneous fistula associated with mediastinitis.

KEYWORDS

case report, post-CABG pericardial effusion, mediastinitis, tracheocutaneous fistula, deep sternal wound infection (DSWI)

Introduction

Post-operative pericardial effusion after cardiac surgery is a relatively common finding, occurring in up to 64% of 780 patients undergoing cardiac surgery in a study by Pepi et al. (1). The majority of cases are asymptomatic and resolve spontaneously. However, up to 1.9% of patients develop features of cardiac tamponade, which occurs more often following valve surgery than coronary artery bypass surgery alone. Another study by Ashikhmina et al., involving 21,416 patients after cardiac surgery, detected symptomatic pericardial effusion in 1.5% of patients (2).

Post-operative pericardial effusion can be classified based on the timing of its development as early vs. late presentation (3). Early presentation pericardial effusion is defined as occurring within the first week of operation and is predominantly related to procedural-related bleeding. Late presentation pericardial effusion occurs after post-operative day 7 and is predominantly inflammatory-mediated. If symptomatic, patients may present with either features of pericarditis, such as pleuritic chest discomfort, or even features of pericardial tamponade, such as tachycardia, breathlessness, and low blood pressure. Patients who present with features of clinical pericardial tamponade would need to be treated with pericardiocentesis, which provides immediate relief of symptoms and hemodynamic stabilization.

Mediastinitis is an infrequent but serious complication that may manifest in the post-operative recovery period. Early detection and treatment can reduce morbidity. Our patient presented with late post-operative pericardial tamponade and improved clinically after pericardiocentesis. During his hospitalization, he did not exhibit any

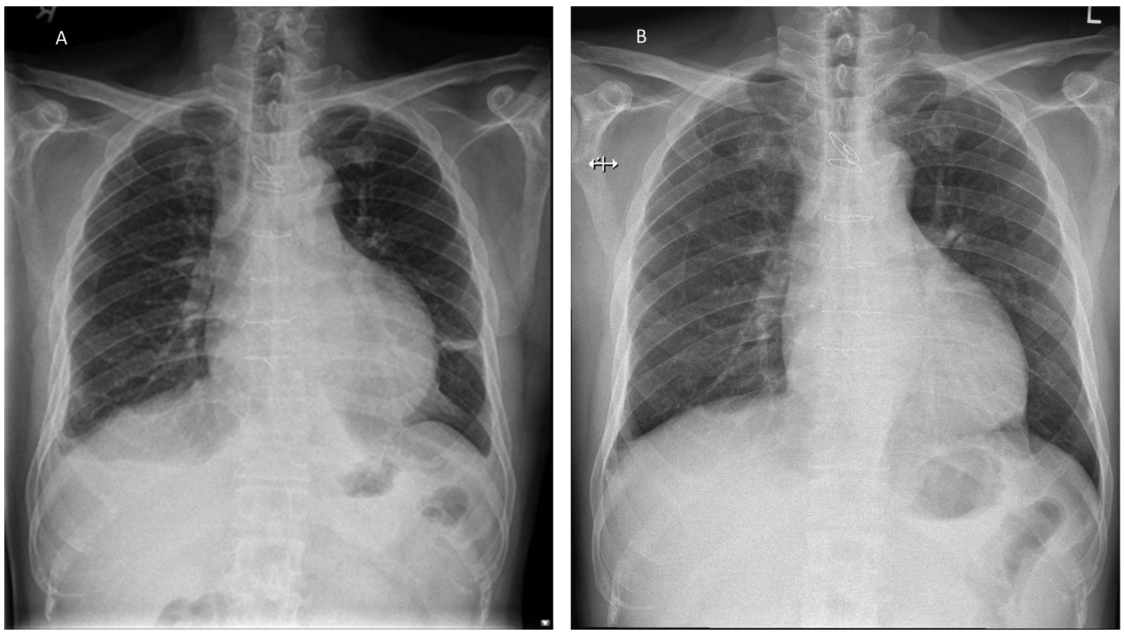

FIGURE 1
Patient's chest x-ray. **(A)** CXR during his admission for pericardial tamponade showing a globular heart and small pleural effusion. **(B)** Outpatient CXR taken 8 days later, showing that the third and fourth sternal wires had fractured with widening of the fifth and sixth wires.

symptoms worrisome for mediastinitis, but unfortunately, he was later readmitted for this complication.

Case description

Our patient is a 62-year-old man with cardiac risk factors of cigarette smoking, hypertension, and hyperlipidemia. He presented with unstable angina and was treated with an early elective coronary artery bypass surgery. His echocardiogram was normal, with a left ventricular ejection fraction of 60% and no pericardial effusion.

Readmission for pericardial tamponade

He was readmitted 11 days later due to breathlessness, orthopnea, and mild pedal edema. He was afebrile, with a blood pressure of 118/79 mmHg and a heart rate of 102 beats per minute. Physical examination revealed muffled heart sounds and mildly elevated jugular venous pulsation. His chest x-ray showed a globular heart with intact sternal wires. Echocardiography showed a large pericardial effusion (measuring 4.1 cm) with features of early cardiac tamponade. His blood results were as follows: hemoglobin (Hb) 11.7 g/dl, white blood cell count (WBC) 13.8×10^9/L, platelet count 397×10^9/L, C-reactive protein (CRP) 13.8 mg/L, procalcitonin < 0.06 µg/L, and creatinine 103 µmol/L.

Progress

He underwent urgent pericardiocentesis, during which 600 mL of hemoserous fluid was drained, resulting in the relief of symptoms. The pericardial drain was inserted in the cardiovascular suite under sterile conditions. No routine antibiotics were given. The pericardial drain was left *in situ* for 2 days and was removed once no further drainage was observed. Microscopic analysis of the fluid drained showed a WBC count of 473/µL, with 75% lymphocytes and 17% polymorphs. Gram staining did not identify any organisms. Aerobic, anaerobic, and acid-fast bacilli cultures were also negative. Fluid analysis showed a fluid lactate dehydrogenase (LDH) level of 384 (serum 408) U/L and a fluid protein level of 57 (serum 76) g/L, fulfilling Light's criteria[1] for an inflammatory effusion. The cytology report showed occasional reactive mesothelial cells admixed with scattered small lymphocytes, some macrophages, and neutrophils in a hemorrhagic background, with no malignant cells identified. The patient remained clinically stable and was discharged on day 3 of admission. He was reviewed at the clinic 5 days later, complaining of a cough with purulent sputum. He was afebrile, and the previous sternotomy and pericardiocentesis sites were clean. A chest x-ray was done, which showed fractured sternal wires (Figure 1).

[1]Light's criteria suggest a fluid is exudative if fluid protein/serum protein ratio >0.5, fluid LDH/serum LDH ratio >0.6, or fluid LDH >2/3 of the upper reference range of serum LDH.

10.3389/fcvm.2025.1483395

Second readmission

The patient was readmitted 2 days later with complaints of worsening cough, blood-tinged sputum, orthopnea, pedal edema, fatigue, and purulent discharge from the upper part of his sternal wound. He was afebrile, with a blood pressure of 84/59 mmHg and a heart rate of 70 beats per minute. Physical examination revealed a small (3 mm) wound over his midline sternotomy that moved with respiration. His heart sounds were normal, but pulmonary crepitations were noted. A bedside echocardiogram showed a moderate-sized pericardial effusion (1.6 cm) without any features of tamponade. Blood test results were as follows: Hb 10.4 g/dL, WBC 14.5×10^9/L, platelet count 99×10^9/L, CRP 313 mg/L, procalcitonin 2.6 µg/L, and creatinine 314 µmol/L. An urgent chest CT showed a tracheal fistula to the subcutaneous sternum along the sternotomy defect and subcutaneous emphysema along the manubrium sternum, posterior sternum, and mediastinum, extending over the right chest wall. A small pericardial effusion and mid- and lower-sternal wire dehiscence were noted. The CT features were suggestive of mediastinitis, no large mediastinal collections were seen (Figure 2).

Further management

The patient was stabilized and underwent sternal reopening, mediastinal washout, and application of a vacuum-assisted closure (VAC) dressing. Intraoperatively, the lower four sternal

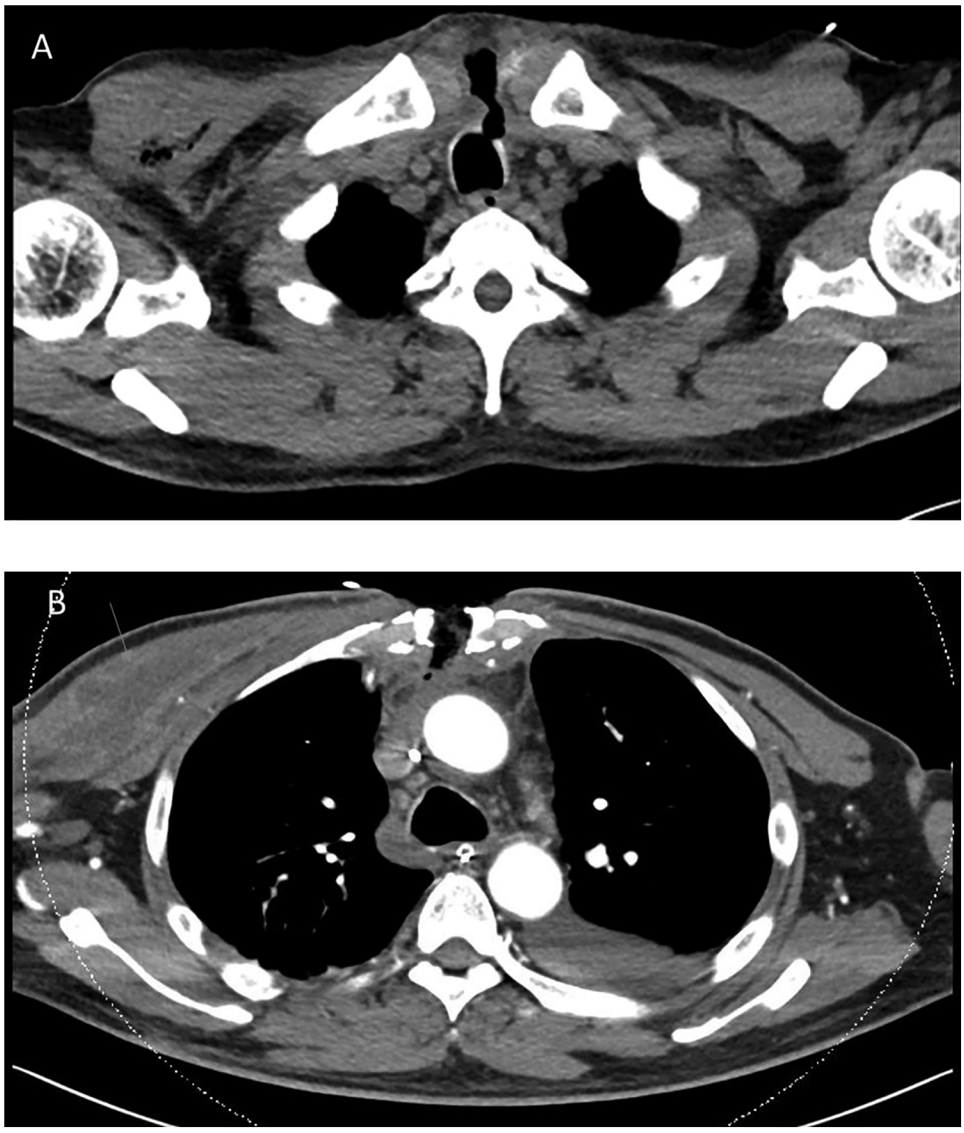

FIGURE 2
Patient's chest CT. **(A)** chest CT performed during his second readmission showing a tracheocutaneous fistula from the upper trachea to the subcutaneous sternum along the sternotomy defect. There was subcutaneous emphysema along the manubrium sternum, posterior sternum, and mediastinum, extending over the right chest wall. No large mediastinal collection was noted in the study. **(B)** Follow-up chest CT performed 1 week later showing interval development of a pectoralis major abscess (arrow), which required further surgical drainage.

wires were noted to be fractured, and unhealthy tissue with areas of fat necrosis was noted over the sternum. However, no frank pus or mediastinal abscess was observed. The following day, a relook surgery was performed, and a temporary seal of the tracheal defect was achieved with a Hemopatch. An interval chest CT at 1 week showed the development of an abscess in the right pectoralis major muscle. He underwent further surgery with an exploration of the neck, repair of the tracheal defect with a sternocleidomastoid muscle flap, debridement of the chest wound, and drainage of the right pectoralis major abscess. He eventually underwent sternal plating and primary closure of the right chest wall wound on post-operative day 9 from the sternal reopening surgery. Wound and sternal bone cultures grew *Klebsiella* and *Streptococcus anginosus*, while blood cultures grew *S. anginosus*. The patient was initially treated with intravenous meropenem and vancomycin before eventually switching to ceftriaxone and metronidazole. He was eventually discharged on post-operative day 17 with oral levofloxacin and metronidazole for an additional 4 weeks. One week after discharge, he was reviewed in the clinic and was noted to have a small area of skin erythema and bogginess (1.5 cm) over the upper part of his chest wound. He was readmitted for sternal wound debridement to the level of the sternal plate, followed by VAC dressing application until wound coverage was achieved 2 weeks later. He remained well during subsequent follow-up visits, and there were no further issues with wound healing or cardiac symptoms (Figure 3).

Discussion

Mediastinitis is a feared complication of coronary artery bypass surgery with significant morbidity and mortality risk. Its incidence is low, with estimates ranging from 0.4% to 4%. In a study by Risenes et al. (4), the incidence of mediastinitis in a cohort of 18,532 patients was 0.6%, with a median presentation of 12 days (range 9–19 days) post-operatively. Risk factors include being a male patient, aged over 70 years, obesity (BMI $\geq$ 30), having received over 10 units of transfusions, diabetes, and chronic obstructive pulmonary disease (COPD). Mediastinitis is associated with significant morbidity, such as prolonged hospital stays, arrhythmia, stroke, and myocardial infarction. In addition, it is associated with reduced long-term survival at 10.3-years (49.5% vs. 71.0% survival). In a study by Milano et al. (5), out of 6,459 patients who underwent coronary artery bypass surgery, 1.3% developed mediastinitis. The 90-day mortality rate for the mediastinitis group was 11.8% vs. 5.5% for those without mediastinitis. An increase in interval mortality between 1 and 2 years post-surgery was also noted, with a rate of 8.1% in the mediastinitis group vs. 2.3% in the non-mediastinitis group.

The cause of post-operative mediastinitis is generally presumed to be either the intraoperative introduction of infection into the mediastinum or the spread of infection from the surgical wounds to the mediastinum (6). However, our patient demonstrated several atypical features. He presented with late pericardial effusion and features of pericardial tamponade. He was clinically not septic and showed resolution of symptoms after pericardiocentesis. Furthermore, his pericardial fluid cultures were negative for bacterial growth, and initial blood investigations did not suggest any signs of an ominous infection. Nevertheless, his white blood cell count and C-reactive protein levels were slightly elevated. Had we monitored these daily, they might have indicated that an infection was brewing, triggering an earlier chest CT. The follow-up chest x—ray, performed 2 days before his second readmission, also showed sternal wire fractures. Sternal wire fractures may be seen before mediastinitis manifests clinically and should prompt further investigations.

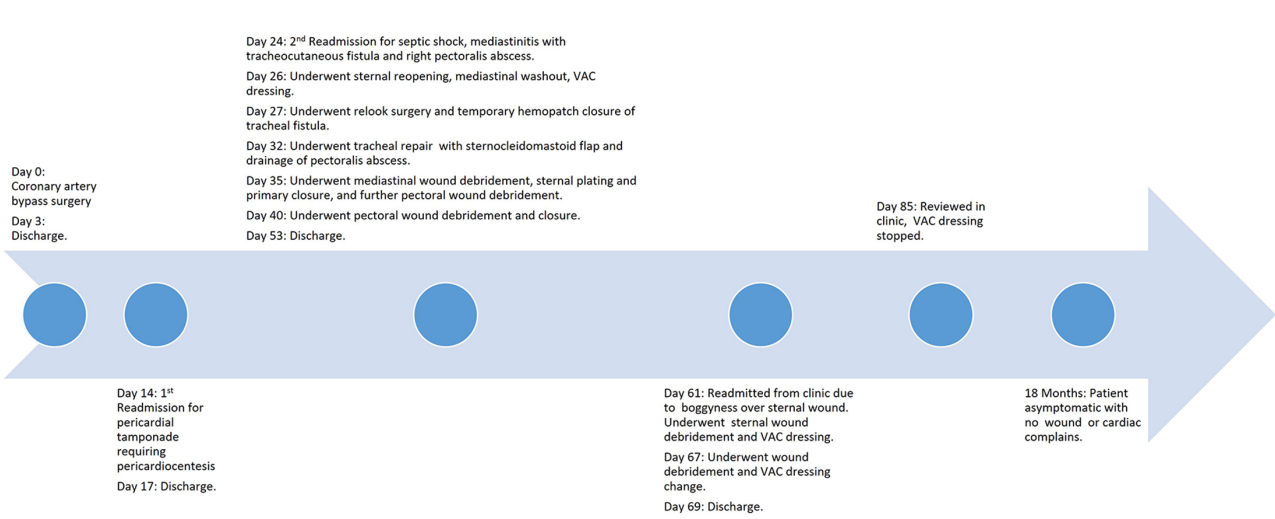

FIGURE 3
Timeline of events.

Tracheomediastinal fistulas are rare and more often seen in patients with cancer (7, 8) or following chemoradiation therapy (9). They have also been described after transbronchial biopsies (10). Routine airway intubation and ventilation have not been shown to cause tracheomediastinal fistulas. The tracheal fistula in our patient was likely from mediastinitis, with tracking of the infection to the trachea and the skin. Although no frank mediastinal abscess was noted on CT scan or intraoperatively, it is possible that an early mediastinal abscess could have drained via the fistula, both into the trachea (accounting for his cough) and subcutaneously to the skin. The infection likely also tracked subcutaneously to the right pectoralis major, where it later formed an abscess collection. In our patient, the most likely cause of his mediastinitis is a post-operative deep sternal wound infection; a less likely differential is bacterial seeding from the pericardiocentesis procedure. The deep sternal wound infection likely caused the mediastinitis and the reactive pericardial effusion.

Due to the significant morbidity and mortality associated with post-operative mediastinitis, delays in diagnosis and treatment should be avoided. As illustrated in this case report, patients may not clinically appear to be septic early on before overt mediastinitis manifests. In the setting of post-operative symptomatic pericardial effusion, a low threshold for CT imaging should be considered, especially if white blood cell count or inflammatory markers remain elevated.

Conclusion

Post-coronary artery bypass surgery pericardial effusions are common and largely asymptomatic. A small minority of patients who present with pericardial tamponade may require pericardiocentesis. While most late-presentation pericardial effusions are reactive and self-resolving, they may be an early manifestation of mediastinitis. Mediastinitis is a major complication associated with significant morbidity and mortality and may present within the first 4 weeks post-operatively. Some patients may initially appear clinically well but may deteriorate rapidly. A high index of suspicion and a low threshold for chest CT should be considered, especially if inflammatory markers are persistently elevated or if sternal wire fractures are noted. Mediastinitis can lead to the rare complication of tracheal fistula.

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

MI: Writing – original draft, Writing – review & editing. CL: Writing – review & editing. KC: Writing – review & editing.

Funding

The author(s) declare that no financial support was received for the research, authorship, and/or publication of this article.

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.

Generative AI statement

The author(s) declare that no Generative AI was used in the creation of this manuscript.

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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.2025.1483395/full#supplementary-material

SUPPLEMENTARY VIDEO 1
Video clip of Chest CT.

References

1. Pepi M, Muratori M, Barbier P, Doria E, Arena V, Berti M, et al. Pericardial effusion after cardiac surgery: incidence, site, size, and haemodynamic consequences. *Br Heart J*. (1994) 72(4):327–31. doi: 10.1136/hrt.72.4.327

2. Ashikhmina EA, Schaff HV, Sinak LJ, Li Z, Dearani JA, Suri RM, et al. Pericardial effusion after cardiac surgery: risk factors, patient profiles, and contemporary management. *Ann Thorac Surg*. (2010) 89(1):112–8. doi: 10.1016/j.athoracsur.2009.09.026

3. Khan NK, Järvelä KM, Loisa EL, Sutinen JA, Laurikka JO, Khan JA. Incidence, presentation and risk factors of late postoperative pericardial effusions requiring invasive treatment after cardiac surgery. *Interact Cardiovasc Thorac Surg*. (2017) 24(6):835–40. doi: 10.1093/icvts/ivx011

4. Risnes I, Abdelnoor M, Almdahl SM, Svennevig JL. Mediastinitis after coronary artery bypass grafting risk factors and long-term survival. *Ann Thorac Surg*. (2010) 89(5):1502–9. doi: 10.1016/j.athoracsur.2010.02.038

5. Milano CA, Kesler K, Archibald N, Sexton DJ, Jones RH. Mediastinitis after coronary artery bypass graft surgery. *Circulation*. (1995) 92(8):2245–51. doi: 10.1161/01.CIR.92.8.2245

6. Kappus S, King O. *Mediastinitis*. Treasure Island, FL: Statpearls.

7. Ucer M, Ordu C, Pilanc KN, Dalar L. Tracheomediastinal fistula in a patient with lung adenocarcinoma and its treatment with argon plasma coagulation: a case report. *Medicine (Baltimore)*. (2014) 93(23):e156. doi: 10.1097/MD.0000000000000156

8. Tse DG, Summers A, Sanger JR, Haasler GB. Surgical treatment of tracheomediastinal fistula from recurrent Hodgkin's lymphoma. *Ann Thorac Surg*. (1999) 67(3):832–4. doi: 10.1016/S0003-4975(98)01342-3

9. McCarthy J, Hamel J. Tracheal-mediastinal fistula post-chemoradiation therapy. *West J Emerg Med*. (2014) 15(7):876–7. doi: 10.5811/westjem.2014.8.23438

10. Jang JG, Ahn JH, Lee SS. Delayed onset of mediastinitis with tracheomediastinal fistula following endobronchial ultrasound-guided transbronchial needle aspiration; A case report. *Thorac Cancer*. (2021) 12(7):1134–6. doi: 10.1111/1759-7714.13888

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TYPE Case Report
PUBLISHED 24 February 2025
DOI 10.3389/fcvm.2025.1480363

OPEN ACCESS

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RECEIVED 13 August 2024
ACCEPTED 06 February 2025
PUBLISHED 24 February 2025

CITATION
Nunez-Ordonez N, Amado-Olivares AF, Jimenez-Ordonez AF, Obando C, Chalela T, Senosiain J, Sandoval N, Camacho-Mackenzie J and Villa-Hincapié C (2025) Case Report: A usual procedure in an unusual situation: a patient with a rare Ehlers Danlos/osteogenesis imperfecta overlap undergoing aortic valve replacement.
Front. Cardiovasc. Med. 12:1480363.
doi: 10.3389/fcvm.2025.1480363

Case Report: A usual procedure in an unusual situation: a patient with a rare Ehlers Danlos/osteogenesis imperfecta overlap undergoing aortic valve replacement

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Connective tissue disorders are known to cause cardiac and vascular complications. We present the case of a 37-year-old female patient with a rare Ehlers Danlos/Osteogenesis Imperfecta Overlap Syndrome, referred to cardiac surgery with aortic valve regurgitation, who underwent a successful Biological Surgical Aortic Valve Replacement (SAVR). A multidisciplinary, patient-centered, heart-team approach is essential in managing patients with rare genetic disorders to optimize postoperative outcomes. Adult cardiac surgeons must become familiar with genetic syndromes and their implications for improving perioperative outcomes.

KEYWORDS

aortic valve replacement, Ehlers-Danlos, osteogenesis imperfecta, biological SAVR, genetic syndrome

1 Introduction

Ehlers-Danlos Syndrome (EDS) encompasses a group of connective tissue disorders related to mutations of genes encoding collagen I (1). Cardiac-valvular EDS is a rare variant that shares some of the most common characteristics of classic EDS (Joint hypermobility, skin hyperextensibility) that often requires heart surgery due to severe valvular compromise (2, 3).

We describe a case of an aortic valve replacement due to severe aortic regurgitation in a young patient with a recently described (1) Osteogenesis imperfecta/Ehlers Danlos overlap syndrome.

2 Case presentation

A 37-year-old female patient was referred for cardiac surgical consultation with an incidental finding of a moderate to severe aortic regurgitation. Upon first consultation

the patient had no symptoms related to cardiac disease. The patient had a prior diagnosis of type I Osteogenesis Imperfecta (OI) at birth that had required multiple interventions due to severe Musculo-skeletal and ocular defects (elbow reconstruction, cornea transplant, spine reconstruction due to severe scoliosis and spondylolisthesis, and ankle osteosynthesis).

2.1 Physical examination

Physical examination revealed a patient with a high palate, an elevated nasal bridge, low-implantation ears, blue sclerae, hypermobility in the fingers and knee joints and hyper-elastic skin. Skeletal deformities consistent with the medical history were evident. Cardiovascular evaluation was positive for a grade III/VI holodiastolic murmur best heard at the right 2nd intercostal space.

2.2 Additional studies

Preoperative echocardiogram (Figure 1a) showed a severely dilated left ventricle with eccentric hypertrophy, LVEF 60%, and severe aortic regurgitation. The aortic root measured 36 mm. Cardiac MRI confirmed the findings with a regurgitant fraction of 59%. The ascending aorta was measured at $30 \times 30 \times 30$ at the highest diameter (commissural level) (Figure 1b). Ergospirometry revealed no limitations in cardiovascular or pulmonary function.

2.3 Genetic evaluation

Given the history of OI, a genetic consultation was requested as part of the workup for Heart Team evaluation. The genetic history revealed one sister with a confirmed diagnosis of type I OI, while no other close relatives had a genetic diagnosis. A genetic sequencing was positive for a heterozygotic mutation of COL1A1 c. 572G>A (p. Gly191Asp.). The final genetic evaluation considered that the patient exhibited findings suggestive of a rare, recently described OI/EDS overlap syndrome: the genetic sequencing showed characteristic glycine residue substitutions on the type I collagen gene and some clinical features (such as blue sclerae, bone fragility or osteopenia) that were compatible with OI; on the other hand, additional findings like the cardiac-valvular compromise, soft-tissue fragility or the joint and skin hypermobility (combined with the identified mutations on the type I collagen gene) suggested the EDS spectrum.

2.4 Procedure

After multidisciplinary discussion and agreement with the patient, an elective aortic valve replacement via median sternotomy was decided. Hyperlaxity of all soft tissues was evident and a severely fragile sternal bone with multiple previous fractures was noted. A conventional technique of arterial cannulation in the distal ascending aorta and venous cannulation through the right atrial appendage were used with a left-cavity venting through the right superior pulmonary vein. TEE was used to assess adequacy of the cannulation before entering on bypass. Notably fragile tissues were encountered on these structures and therefore minimal traction and tissue manipulation were sought. A tricuspid aortic valve was found, exhibiting a severe retraction of the non-coronary cusp (Figure 2c). The aortic root and ascending aorta were not dilated. Coronary ostia showed usual disposition. A biological *Edwards Lifesciences INSPIRIS RESILIA* 25 mm aortic valve was implanted without complications, and intraoperative TEE confirmed an adequate functioning valve with normal biventricular systolic function. Special care was taken during sternal closure due to severe frailty of the tissues. A sternal hemi-cerclage was performed, and 6 surgical steel simple sutures were placed to prevent sternal instability or dehiscence (Figure 1c). Clamp time was 72 min, and CPB time was 91 min.

Postoperative recovery was uneventful, early extubation upon arrival to the ICU was achieved. The patient was taken to the general ward after 3 days of ICU monitoring and was discharged home on POD6. Early postoperative TTE indicated a normofunctional prosthesis (peak vel. 1, 8 m/s, peak gradient 12 mmHg, mean gradient 6 mmHg).

Pathological examination of the excised tissue revealed non-specific degenerative changes in the valve stroma with mucin deposits above the valvular tissue (Figures 2a,b).

2.5 Follow-up

At the 12 month postoperative evaluation the patient was asymptomatic, with no signs of postoperative complications; no wound or sternal complications were noted, and a normofunctional valve with normal LV function on 3-month control TTE was confirmed.

3 Discussion

With recent advances in diagnostic and therapeutic techniques, patients with diverse genetic conditions have become increasingly common in adult cardiac surgery practice. It is therefore important for adult cardiac surgeons worldwide to become familiar with the challenges that each condition may impose on otherwise common surgical procedures.

We presented a case of a patient with a rare OI/EDS overlap. This is, to the author's knowledge, the first reported experience of a patient with such condition undergoing cardiac surgery. We provide an overview on the relevant characteristics of this COL-1

Abbreviations

EDS, Ehlers Danlos syndrome; ICU, intensive care unit; LVEF, left ventricular ejection fraction; MRI, magnetic resonance imaging; OI, osteogenesis imperfecta; POD, post - operative day; SAVR, surgical aortic valve replacement; TEE, transesophageal echocardiogram; TTE, transthoracic echocardiogram.

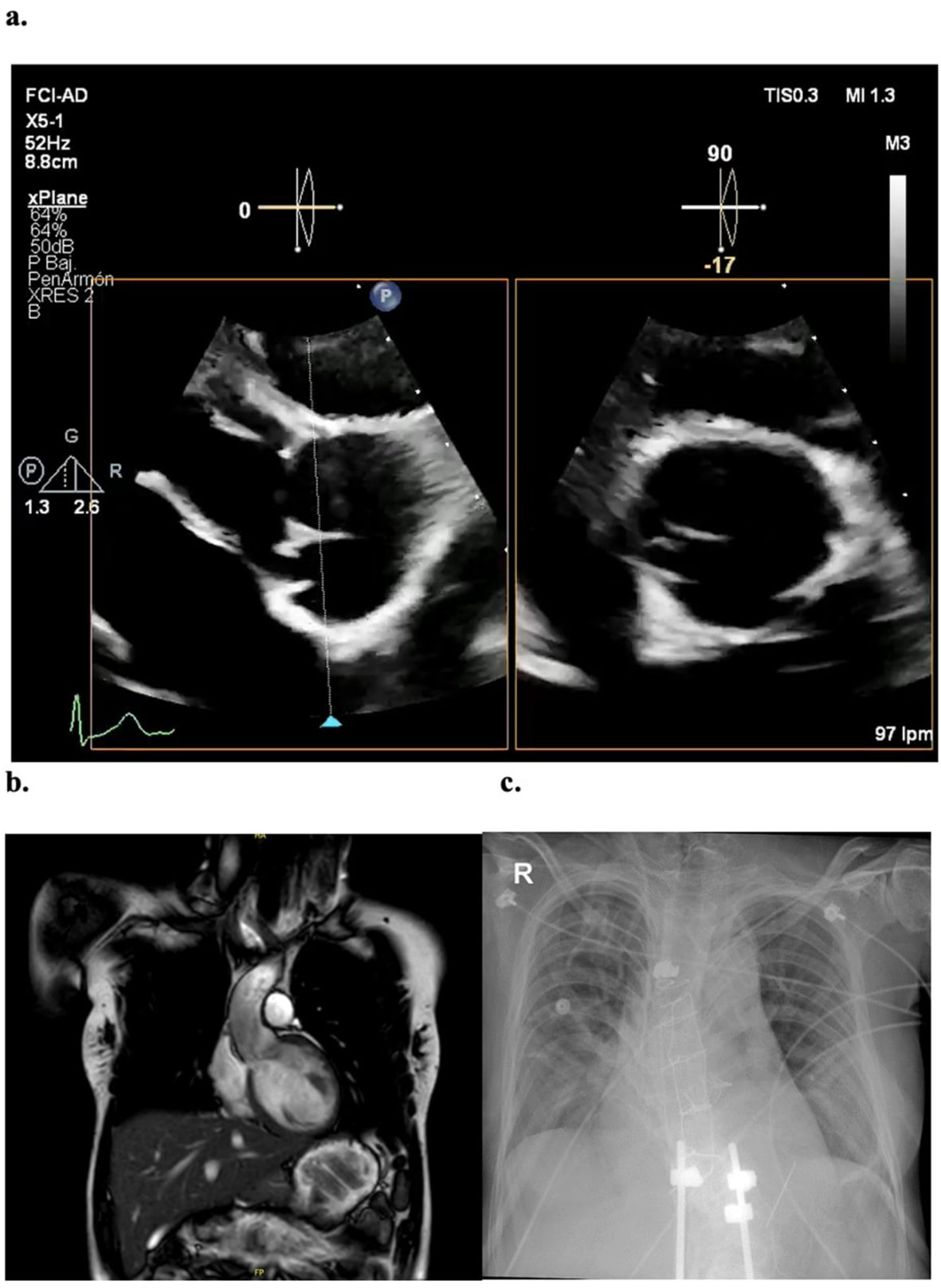

FIGURE 1
Imaging studies (a) preoperative transthoracic echocardiogram, with severe aortic regurgitation associated to a non-coronary cusp retraction. (b) Coronal view of Cardiac MRI, showing a non-dilated ascending aorta with maximum commissural diameter of 30 mm (c) postoperative x-ray evidencing 6 surgical steel wire sutures and sternal hemi cerclage.

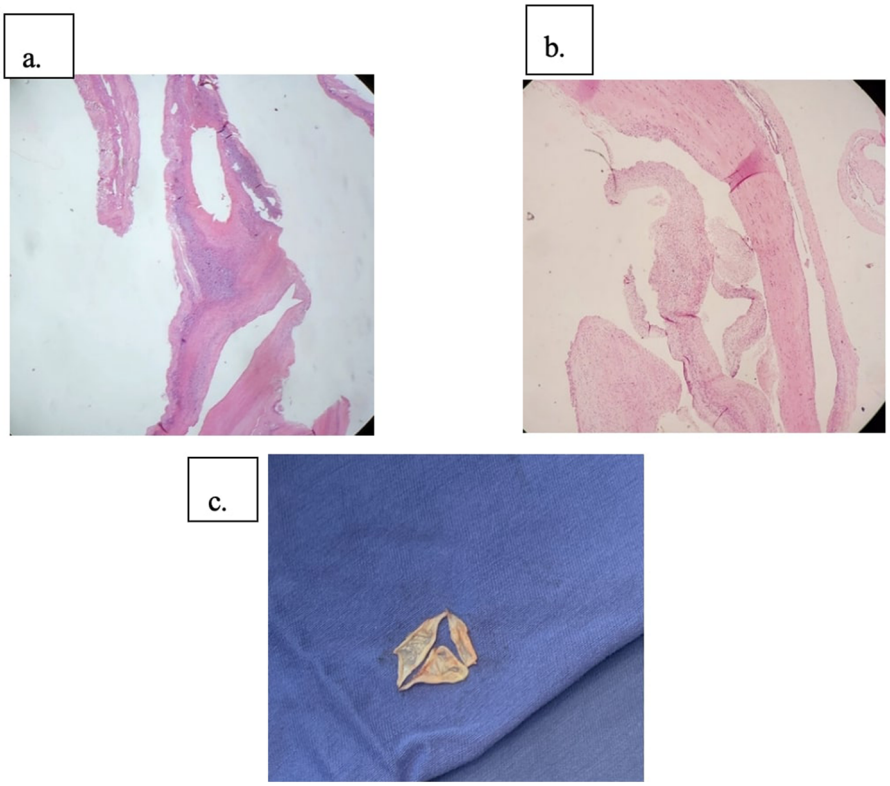

FIGURE 2
Pathologic findings **(a)** hematoxylin eosin stain showing degenerative changes in valvular stroma. **(b)** Alcian blue stain, blue-stained areas that correspond to mucin deposits **(c)** surgical specimen showing a tricuspid aortic valve with retraction of the non-coronary cusp.

related disorder for the cardiac surgeon and some insights on the perioperative considerations to improve outcomes.

3.1 The syndrome

EDS refers broadly to a group of connective tissue disorders related mainly to mutations of the genes encoding collagen (2), manifesting with joint hypermobility, skin hyperextensibility and soft tissue and vascular fragility (1). Patients with COL1A2 mutations result in absent/reduced proα2 chains with a resultant inability to adequately produce the protein. Cardiac-valvular EDS is a rare variant of the disease (4). Patients with Cardiac-valvular EDS present with classical EDS features such as joint hypermobility, skin hyperextensibility while also developing cardiac valvular disease. These patients frequently develop mitral valve disease (usually mitral valve prolapse) but aortic valve compromise is not uncommon (2).

Osteogenesis imperfecta, on the other hand, is an autosomal dominant disease (5) that results from quantitative or qualitative defects in type I collagen. Most mutations are related to COL1A1 and COL1A2 genes. Patients frequently present with bone fragility, blue sclera, dental fragility, hearing abnormalities and hyperlaxity. Type I collagen is also a structural component in cardiac and vascular extracellular matrix making these patients prone to cardiovascular manifestations (although infrequent). Among those, aortic regurgitation due to leaflet dysfunction as in this case, has been reported (6, 7), but mitral compromise seems to be more common.

A recently described COL1-related disorder manifesting as an OI/EDS overlap (1) was diagnosed in this patient. This is an extremely rare combination reaching an incidence around 1/1,000.000 (8). Even if proper diagnostic criteria have not been established so far, the phenotype of this patient is similar to what has been reported elsewhere (8). A characteristic glycine substitution was identified in this case which could be responsible for the clinical presentation.

3.2 Surgical considerations

As seen in this case, patients with collagen-related disorders also require common cardiac surgical interventions, such as valvular replacement surgery or aortic root/ascending aorta surgery. However, several critical concerns are worth highlighting from this case: major issues were anticipated with handling of the weak, friable tissues; a high intra- and postoperative bleeding risk was recognized, and difficulties with sternal opening and closure were expected as well. Other potential complications may be related to a higher risk of perioperative arrhythmias due to a possible involvement of the cardiac conduction system as a part of the syndrome (6). Long-term risk such as valve dehiscence or

aortic dissection should be kept into account however, a careful surgical procedure may mitigate these concerns.

Patients with collagen-related disorders have high reported mortality rates. One recent study reported a perioperative mortality exceeding 20%, mainly related to bleeding complications (6). Therefore, additional measures must be taken when caring for these types of patients.

Adequate surgical planning should include the selection of the optimal timing for the intervention, while minimizing the presence of other risk factors. In this case, the patient was being followed as an outpatient and surgery was decided in the presence of mild symptoms and guided primarily by imaging criteria given that it was considered the best scenario for a safe surgical procedure as the patient was in an overall good general health condition.

3.3 Intraoperative considerations

Multiple special concerns should be considered when taking care of patients with COL1-related disorders. From anesthetic care (including the risk of temporo-mandibular joint dislocation) to a higher chance of organ rupture or inducing vessel injuries with surgical handling, these patients need special attention during the intraoperative period (4, 6). Surgical planning incorporates all these aspects and raising awareness on the whole surgical team on these multiple issues is essential.

Gentle tissue handling and even including less invasive approaches (such as mini sternotomies) is recommended for these procedures (9). In this case, however, a full sternotomy was performed due to the treating surgeon's preference as it was believed that the conventional cannulation and surgical technique were safer and effective while reducing the risk of retrograde aortic dissection associated with femoral cannulation. Our approach included a preoperatively planned preventive sternal cerclage with surgical steel which proved effective in preventing sternal dehiscence at 12 months.

3.4 Ideal valvular prosthesis

Choosing the most appropriate valvular prosthesis is a critical decision for these patients. Current guidelines by the ACC/AHA recommend considering a mechanic aortic prosthesis in patients younger than 50 years of age (class of recommendation 2a) (10). However, in accordance with both the AHA/ACC and ESC/EACTS current guidelines (10, 11), this patient was deemed at high bleeding risk due to comorbidities; therefore, a bioprosthesis was chosen. Additionally, considering the patient's medical history, it was likely that future surgical interventions (both cardiac or non-cardiac) would be necessary, making the bioprosthesis a risk-reducing option.

A recent review by Dimitrakakis et al. on the ideal prosthesis for patients with osteogenesis imperfecta showed that bioprosthetic valves tend to yield better outcomes as compared to mechanical valves. Since these patients tend to have a high bleeding risk due to friable tissues, platelet dysfunction and capillary fragility, bioprosthetic valves seem to be a reasonable choice in terms of controlling bleeding complications. Besides, the implantation of mechanical valves may generate more mechanical trauma to the weakened, friable tissues, thus increasing the risk of aortic dissection and paravalvular leaks (5).

3.5 Postoperative care

In this case routine postoperative care was provided and guaranteed an uneventful recovery. Special care and awareness should be kept on the perioperative bleeding risk.

Another concern that should guide postoperative care is a delayed wound healing that is frequent in this subset of patients. From soft tissue to sternal healing, providers should be aware and anticipate the possible wound complications that could develop in the postoperative course.

3.6 Take-away lessons

A multidisciplinary, patient-centered, heart-team approach provides is fundamental in decision making related to perioperative care of patients with rare genetic disorders for improving outcomes.

Advances in diagnostic and therapeutic methods has led to an increase in patients with diverse genetic disorders that undergo cardiac surgery, challenging adult cardiac surgeons to become familiar with characteristics of each syndrome and its implications for improving perioperative outcomes.

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 humans were approved by Comite de Etica en Investigacion - Fundacion Cardioinfantil-LaCardio. The studies were conducted in accordance with the local legislation and institutional requirements. The 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

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Funding

The author(s) declare that no financial support was received for the research, authorship, and/or publication of this article.

Acknowledgments

To the multidisciplinary team at Fundación Cardioinfantil/LaCardio allowing excellent patient-centered care.

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

References

1. Morlino S, Micale L, Ritelli M, et al. COL1-related overlap disorder: a novel connective tissue disorder incorporating the osteogenesis imperfecta/Ehlers-Danlos syndrome overlap. *Clin Genet*. (2020) 97(3):396–406. doi: 10.1111/cge.13683

2. Brady AF, Demirdas S, Fournel-Gigleux S, et al. The Ehlers-Danlos syndromes, rare types. *Am J Med Genet C Semin Med Genet*. (2017) 175(1):70–115. doi: 10.1002/ajmg.c.31550

3. Guarnieri V, Morlino S, Di Stolfo G, Mastroianno S, Mazza T, Castori M. Cardiac valvular Ehlers-Danlos syndrome is a well-defined condition due to recessive null variants in COL1A2. *Am J Med Genet A*. (2019) 179(5):846–51. doi: 10.1002/ajmg.a.61100

4. Azanza DXC, Munín MA, Sánchez GA, Spernanzoni F. Prolapse and regurgitation of the four heart valves in a patient with Ehlers-Danlos syndrome: a case report. *Eur Heart J Case Rep*. (2019) 3(2):1–8. doi: 10.1093/EHJCR/YTZ052

5. Dimitrakakis G, Challoumas D, Von Oppell UO. What type of valve is most appropriate for osteogenesis imperfecta patients? *Interact Cardiovasc Thorac Surg*. (2014) 19(3):499–504. doi: 10.1093/icvts/ivu152

6. Lamanna A, Fayers T, Clarke S, Parsonage W. Valvular and aortic diseases in osteogenesis imperfecta. *Heart Lung Circ*. (2013) 22(10):801–10. doi: 10.1016/j.hlc.2013.05.640

7. Bonita RE, Cohen IS, Berko BA. Valvular heart disease in osteogenesis imperfecta: presentation of a case and review of the literature. *Echocardiography*. (2010) 27(1):69–73. doi: 10.1111/j.1540-8175.2009.00973.x

8. Morabito LA, Allegri AEM, Capra AP, et al. Osteogenesis Imperfecta/Ehlers-Danlos overlap syndrome and neuroblastoma-case report and review of literature. *Genes (Basel)*. (2022) 13(4):581. doi: 10.3390/genes13040581

9. Izzat MB, Wan S, Wan IYP, Khaw KS, Yim APC. Ministernotomy for aortic valve replacement in a patient with osteogenesis imperfecta. *Ann Thorac Surg*. (1999) 67(4):1171–3. doi: 10.1016/S0003-4975(99)00126-5

10. Otto CM, Nishimura RA, Bonow RO, 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. *Circulation*. (2021) 143(5):E72–227. doi: 10.1161/CIR.0000000000000923

11. Vahanian A, Beyersdorf F, Praz F, 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

frontiers | Frontiers in Cardiovascular Medicine

TYPE Case Report
PUBLISHED 25 February 2025
DOI 10.3389/fcvm.2025.1549613

OPEN ACCESS

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RECEIVED 21 December 2024
ACCEPTED 11 February 2025
PUBLISHED 25 February 2025

CITATION
Peng D, Xu G, Wang B, Li X and Wang Y (2025) Aortic endograft infection after thoracic endovascular aortic repair: two case reports and literature review.
Front. Cardiovasc. Med. 12:1549613.
doi: 10.3389/fcvm.2025.1549613

Aortic endograft infection after thoracic endovascular aortic repair: two case reports and literature review

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With the maturity of thoracic endovascular aortic repair (TEVAR) technology and its increasing application in clinical practice, complications and long-term management after TEVAR have become issues of concern. Here, we report two cases of TEVAR for thoracic aortic dissection. One patient developed recurrent fever 6 years after TEVAR and underwent multiple courses of antibiotic therapy with a poor response. He came to our hospital 6 months later and presented with gastrointestinal bleeding. Imaging revealed the presence of an aortic abscess around the stent graft involving the esophagus and mediastinum. The patient's condition deteriorated rapidly after admission, and he ultimately succumbed to hemorrhagic shock. Another patient developed recurrent fever 1 year after surgery. Imaging studies suggested an aortic abscess with involvement of the esophagus, and the patient chose conservative treatment. After long-term anti-infective treatment, the infected lesions remained but had decreased in size. Aortic endograft infection complicated by multiple organ involvement is a rare complication of TEVAR and has a high mortality rate. After an extended postsurgical period, patients who have undergone TEVAR often lack regular follow-up and are easily overlooked. Our cases highlight the importance of early prevention, early diagnosis, and appropriate management of late complications following TEVAR.

KEYWORDS

TEVAR, complications, long-term management, aortic endograft infection, aortoesophageal fistula, gastrointestinal bleeding

Introduction

Diseases of the aorta, including thoracic aortic aneurysm (TAA) and type B aortic dissection (TBAD), have become life-threatening emergencies in some acute complex conditions. With the development of endovascular techniques, surgical treatment for thoracic aortic emergencies has shifted from traditional open surgery to minimally invasive thoracic endovascular aortic repair (TEVAR). TEVAR was recommended by the European Society for Vascular Surgery (ESVS) as a first-line treatment for descending aortic rupture, penetrating aortic ulcers, intramural hematoma, blunt traumatic aortic injury, and complicated acute type B aortic dissection in 2017 (1). Although TEVAR has achieved impressive therapeutic effects and has successfully saved the lives of countless patients, complications related to stent placement should not be overlooked. Here, we report two cases of patients who developed severe complications 1 and 6 years after TEVAR. Both of these patients developed an aortic abscess around the stent graft involving the esophagus, and ultimately, one of the patients died.

Currently, sufficient academic literature on the long-term outcomes of TEVAR treatment are lacking. Our case report aims to provide a reference for the prevention and diagnosis of long-term complications after TEVAR.

Case report 1

A 59-year-old male patient underwent TEVAR surgery 6 years prior due to thoracic aortic dissection. Postoperatively, he regularly took metoprolol and irbesartan to control his blood pressure, and his condition remained stable. Six months prior, the patient developed irregular fever without apparent cause, with a maximum temperature of 39.5°C. He also experienced profuse sweating and fatigue. At a local hospital, chest CT and echocardiography showed no abnormalities. Cephalosporins and penicillins were administered for more than 20 days, resulting in the relief of fever symptoms. However, fever recurred after discharge, and subsequent antibiotic treatment at a local clinic showed poor efficacy. One month prior, the patient developed cough with the expectoration of small amounts of white sputum but did not receive standardized treatment. Five days prior, he experienced increased urinary frequency, urgency, and dysuria. On the morning of admission, the patient experienced hematemesis, characterized by bright red blood in small amounts. He presented to our hospital for evaluation. Over the past 6 months, the cause of his fever has remained unknown, and he has lost 10 kg in weight.

At admission, his physical examination results were as follows: poor general condition, blood pressure, 108/70 mmHg; temperature, 36.4°C; heart rate, 98 beats/min; and respiratory rate, 20 breaths/min. He exhibited epigastric tenderness without abdominal tension or rebound tenderness. Other vital signs are unremarkable.

The laboratory test results were as follows: white blood cell count, 20.5×10^9/L (normal range: 3.5–9.5); neutrophil percentage (NE%), 0.88 (normal range: 0.40–0.75); absolute neutrophil count, 18.1×10^9/L (normal range: 1.8–6.3); and hemoglobin, 112 (normal range: 130–175) g/L. His inflammatory markers were elevated: C-reactive protein was 256.93 (normal range: 0–10) mg/L, procalcitonin was 24.73 (normal range: 0–0.5) ng/ml, the erythrocyte sedimentation rate was 43 (normal range: 0–15) mm/h. blood culture (aerobic + anaerobic) revealed *Streptococcus constellatus* (anaerobic).

Although the patient's chest CT and echocardiogram at other hospitals showed no abnormalities, an infection related to TEVAR could not be excluded. After admission, antibiotic therapy was adjusted to meropenem [1.0, ter in die, intravenous drip (TID, iv. D)] combined with tigecycline [0.05, bis in die, intravenous drip (BID, iv. D)] for infection control. Eight hours after admission, the patient experienced dizziness and passed 400 ml of bloody stools. Emergency endoscopy revealed massive gastric hemorrhage, however, the bleeding source could not be identified, and the procedure was terminated. Subsequent thoracic aortic computed tomography angiography (CTA) suggested postthoracic aortic stent grafting, with thrombosis around the stent and the formation of a local fistula. There were low-density shadows and scattered gas shadows outside the stent, suggesting possible abscess formation. The boundary between the stent and the esophagus and lung tissues was unclear, with haziness in the mediastinal fat gap and a slight accumulation of air. There was also enlargement of the mediastinal and left hilar lymph nodes (Figure 1). Considering the patient's history of stent implantation and clinical presentation, the possibility of periaortic infection combined aortoesophageal fistula, gastrointestinal bleeding, endoleak and mediastinal abscess were considered. Oral intake was prohibited, and omeprazole and octreotide were administered continuously.

Following a comprehensive evaluation by the cardiac surgery team, repeat stenting or surgical intervention was deemed not feasible. Twenty-four hours after admission, the patient's blood pressure progressively decreased to 87/51 mmHg, and his hemoglobin decreased to 66 g/L. Continuous infusion of adrenaline was initiated, and aggressive fluid resuscitation and blood transfusion were performed. However, the patient experienced recurrent hematemesis and melena. Eventually, he died of hemorrhagic shock.

Case report 2

A 69-year-old male patient underwent TEVAR surgery 1 year prior due to aortic dissection. Postoperatively, he regularly took amlodipine besylate to control his blood pressure. Additionally, he had a 10-year history of diabetes with poor glycemic control. Six months prior, the patient developed fever without apparent cause, with a maximum temperature of 39.5°C and chills, and blood culture revealed Streptococcus intermedius. He was diagnosed with sepsis at our hospital and was discharged after anti-infective treatment. Two months prior, fever and chills recurred, and the patient was admitted to our hospital. CTA of the thoracic aorta revealed a mass lesion around the stent in the lower thoracic aorta with air accumulation. Aortic endograft infection was considered, and the patient chose conservative treatment. He was discharged after anti-infective treatment with cefotidine. Seven days prior, the patient developed fever with cough and expectoration again. Lung CT at our hospital revealed aggravated bilateral pneumonia, bilateral pleural effusion and a lower esophageal mass. He presented to our hospital for further evaluation.

On admission, his physical examination results were as follows: anemic appearance; blood pressure, 152/78 mmHg; and body temperature, 36.4°C. His heart rate was 78 beats/min, and his respiration rate was 20 beats/min.

The laboratory findings were as follows: percentage of neutrophils (NE%), 0.81 (normal range: 0.40–0.75), absolute neutrophil count, 7.35×10^9/L (normal range: 1.8–6.3); and hemoglobin level, 70 (normal range: 130–175) g/L. His inflammatory marker levels were elevated: C-reactive protein, 69.68 (normal range: 0–10) mg/L and erythrocyte sedimentation rate, 80 (normal range: 0–15) mm/h. Blood cultures were negative.

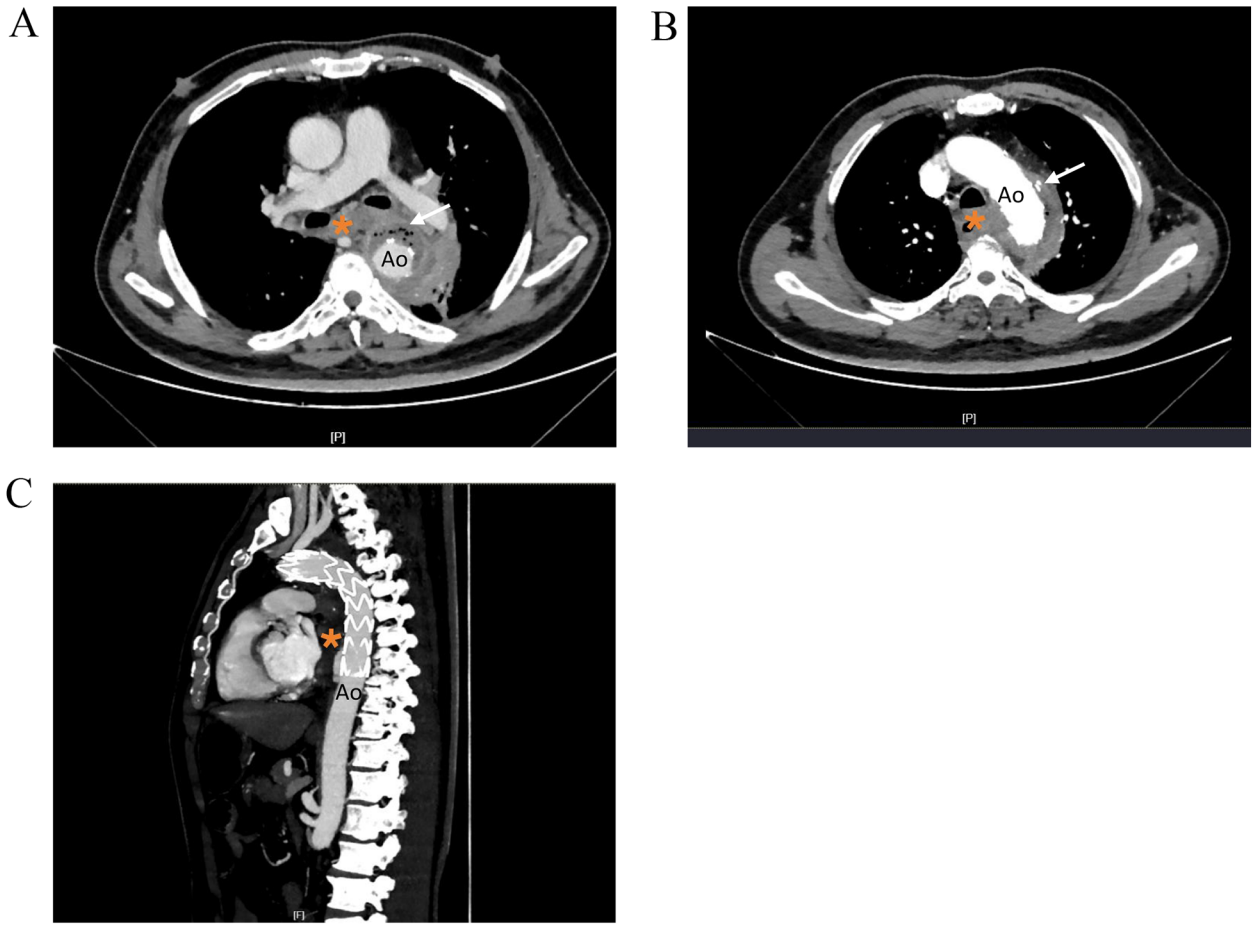

FIGURE 1
Thoracic aortic CTA images showing. **(A)** Low density shadow and scattered gas shadow around the aortic stent graf. **(B)** Nodular contrast imaging was observed outside the stent. **(C)** Sagittal reformatted images. Ao, descending thoracic aorta; *, esophagus.

The patient had recurrent fever for 6 months, After the second admission, and CTA still revealed a mass lesion around the stent. Aortic endograft infection was considered. piperacillin-tazobactam [4.5, ter in die, intravenous drip (TID, iv. D)] and tigecycline [0.05, bis in die, intravenous drip (BID, iv. D)] were given for anti-infective treatment. CTA imaging of the thoracic aorta revealed that after thoracic aortic stenting, there was a mass lesion around the lower thoracic aortic stent and the esophageal area with air accumulation. Compared with the previous CTA of the thoracic aorta, the lesion was enlarged in the right anterior area and slightly reduced in the left posterior area, with bilateral pleural effusions and partial atelectasis in the adjacent lower lobe of the left lung. After anti-infective treatment, the patient had no fever, and the infection index decreased. After multidisciplinary consultation with the Departments of Cardiac Surgery, Vascular Surgery, and Infectious Disease, it was considered that the patient had an aortic graft infection involving the esophagus and that surgical treatment was needed; however, the risk of surgery was high, and the patient chose to continue conservative treatment. Minocycline [0.1, bis in die, per os (BID, p.o.)] and amoxicillin [0.5, ter in die, per os (TID, p.o.)] were recommended after discharge. The patient returned to the hospital for reexamination 3 months after discharge, and there was no fever or other symptoms of discomfort. CTA of the thoracic aorta revealed a mass lesion around the stent in the lower thoracic aorta and the esophageal area with air accumulation, although it was significantly smaller than that previously observed (Figure 2).

Discussion

We report two elderly male patients who developed graft infection following TEVAR. two of our patients developed aortic graft infection involving multiple organs 6 years and 1 year after TEVAR, which is extremely rare and concerning.

The incidence of aortic diseases is increasing annually, and TEVAR technology is increasingly being applied in clinical practice (2). According to a recent publication based on data from the International Registry of Aortic Dissection (IRAD), from 1996 to 2022, the proportion of acute TBAD patients

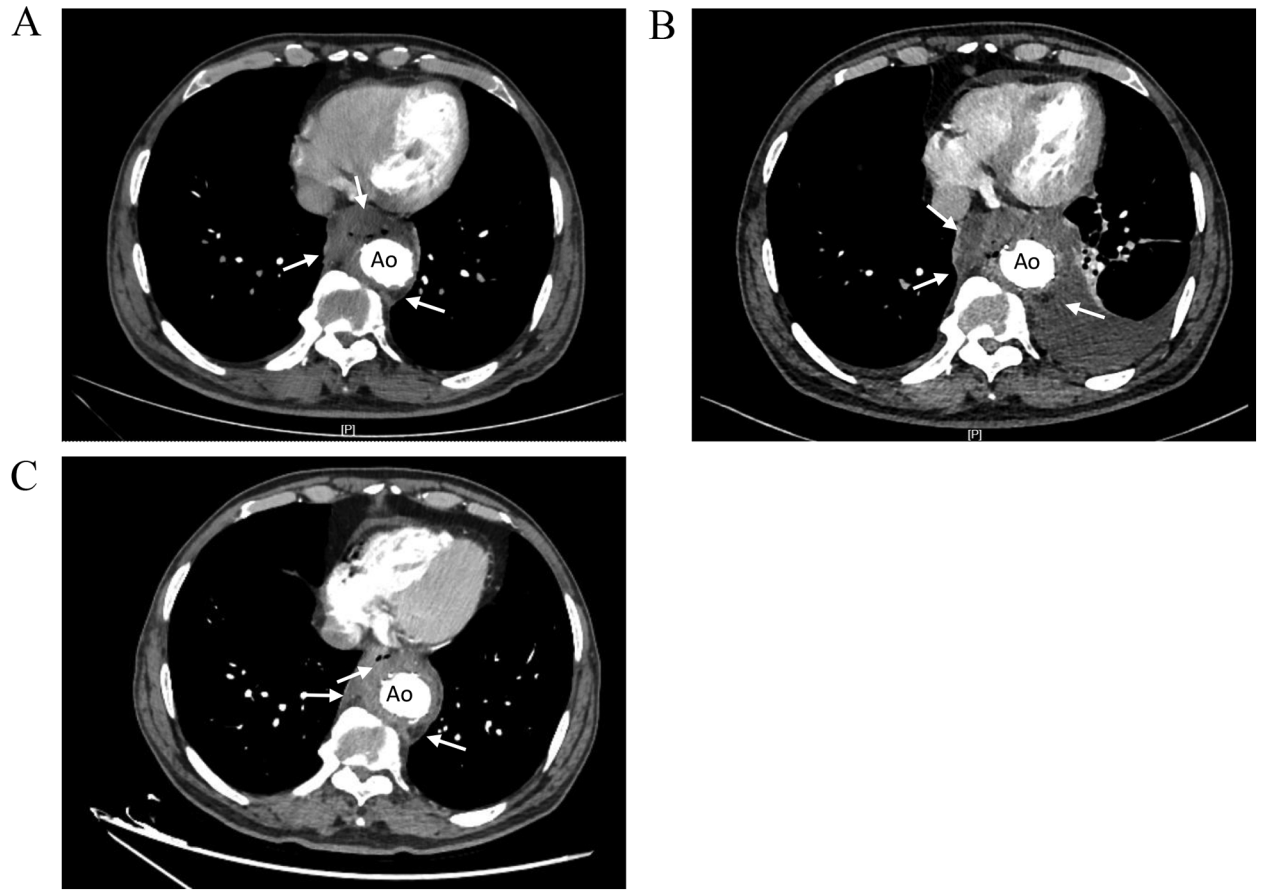

FIGURE 2
Comparison of thoracic aortic CTA images showing. **(A)** CTA images 2 months before admission. Low-density shadow was found around the stent in the lower thoracic aorta (right side, right anterior, left posterior), and the boundary with the esophagus was not clear. **(B)** CTA images during hospitalization. The low-density shadow around the stent in the lower thoracic aorta had unclear boundary with the esophagus, accompanied by air in it, and the scope of the lesion was larger than before. **(C)** CTA images at 3 months after discharge. The mass lesion around the lower thoracic aortic stent with air accumulation in it, the range of infection was smaller than before. Ao, descending thoracic aorta.

receiving endovascular management increased from 19.1% to 37.2% (3). Clinical studies have shown that compared to open aortic repair, TEVAR results in shorter hospital stays and fewer complications (4). Many patients with aortic diseases significantly benefit from this surgical approach. With the increasing use of endovascular aneurysm repair (EVAR), related complications have also increased (5). Some complications after TEVAR, such as endoleak, paraplegia, and stroke, have been widely reported, and some rare complications include aortoesophageal fistula, endograft collapse, and cerebrovascular events (6–8). Although their incidence is low, they often develop in a malignant manner, posing a threat to the patient's life and requiring close attention. Table 1 summarizes some complications of TEVAR, along with their diagnosis and treatment.

In our study, the patients experienced recurrent fever, elevated inflammatory marker levels, and an evident aortic abscess around the stent combined with aortic-esophageal fistula. According to the diagnostic criteria for aortic graft infection (AGI) established by the Management of Aortic Graft Infection Collaboration (MAGIC) in the UK (22), the patient received a definite diagnosis of post-TEVAR AGI. AGI is a rare complication of TEVAR, and its diagnosis and management are highly complex. The clinical presentations are diverse, which is unfavorable for disease diagnosis and treatment.

AGI has diverse causes, and based on the time of occurrence, AGI can be classified into early (<4 months) and late (≥4 months) infections (23). Early AGI is typically caused by contamination during EVAR surgery or a preexisting bacterial infection present during EVAR (24). Late AGI can be influenced by a wide range of factors, and any adjacent or distant infection from various sites can be a causative or contributing factor to AGI. For example, bacteremia resulting from urinary tract or respiratory tract infections can lead to bacterial dissemination via the bloodstream to the area around the graft, causing infection (25). Mechanical erosion due to stent displacement, repetitive frictional tears, and intramural fixation hooks can also lead to fistula formation and inflammation (26). Additionally, researchers have found that factors such as the use of immunosuppressive agents by the host or the presence of other conditions, such as malignancies or diabetes, are significant risk

TABLE 1 Complications, clinical presentations, and treatment after TEVAR.

Complications	Clinical manifestation	Treatment	References
Endoleaks	Imaging reveals persistent blood flow perfusion within the residual aneurysmal sac.	Surgical re-intervention can be performed based on the classification of endoleaks, such as endovascular extension grafts, coil embolization, and conversion to open surgery.	(2, 9, 10)
Stent migration	Displacement of the endoprosthesis from its original position by more than 10 mm is observed or any amount of migration resulting in symptoms.	Using an aortic extension cuff or deploying a large balloon to expand the stent can enhance the fixation of the endoprosthesis to the native aortic wall.	(11, 12)
Endograft collapse	Symptoms suggestive of acute aortic occlusion may arise, with imaging revealing stenosis within the graft.	Balloon dilation can be performed, and open aortic repair may be necessary if needed.	(7, 13)
Spinal cord ischemia	Manifests as permanent or transient spinal cord injury.	Preoperative spinal drainage, perioperative epidural corticosteroid use, and spinal drainage.	(14, 15)
Cerebrovascularevents	Some patients present with cerebral ischemia or asymptomatic cerebral infarction.	Follow the standard stroke management guidelines.	(16, 17)
Post-implantation syndrome	Non-infectious persistent fever and elevated inflammatory markers.	Monitor and manage symptomatically; antibiotics are not required.	(18)
Endograft infection	It often presents with fever, back pain, and elevated inflammatory markers. Computed tomography scans show bubbles or fluid around the aortic graft.	Aggressive antimicrobial therapy; Surgical removal of infected endograft.	(19)
Aortoesophageal fistula	Fever, fatigue, occasionally accompanied by sepsis, clear visualization of fistula on endoscopy.	Aggressive antimicrobial therapy, surgical treatment typically involves a combination of aortic procedures (TEVAR, graft replacement, or repair) and esophageal procedures (esophagectomy, esophageal stenting, or repair).	(20, 21)

factors for AGI (25, 27). In case report 2, the patient had diabetes mellitus and usually had poor glycemic control, which may have increased his risk for infection. Both patients had pulmonary infections during the course of their disease, which also raises the possibility of bacteremia secondary to a respiratory tract infection.

AGI poses catastrophic hazards to patients, as the progression of infection may involve other organs, including the esophagus, trachea, lungs, and mediastinum, leading to complications such as aortoesophageal fistula, aortobronchial fistula, and complex systemic infections (28). The fundamental principles of managing AGI include removing the infected device, performing vascular reconstruction, and administering adjunctive antimicrobial therapy (19). However, vascular grafts are not designed for easy removal, and the surgical procedure imposes significant trauma on patients. One study showed that the 30-day mortality rates for endovascular and open reintervention for failed TEVAR were 6.7% and 15%, respectively (29). For patients who are not candidates for surgery, such as those with a high anticipated surgical mortality, those whose grafts are situated in a location that cannot be resected, or those who decline surgical intervention, antibiotic therapy may present a more favorable alternative to surgical resection. Relevant studies have found that some patients who are inoperable and treated with antibiotics alone have achieved favorable treatment outcomes through long-term, aggressive targeted antibiotic therapy (30, 31). Early targeted antibiotic treatment plays a significant role in eradicating pathogenic microorganisms and reducing the formation of biofilms, among other important effects (30). However, studies have also shown that conservative treatment is difficult to prevent the progression of this disease, and the mortality rate is extremely high (19, 25). One patient in our study was treated conservatively, and the infection lesion was reduced after active anti-infective treatment; however, the infection lesion remained. We will continue to follow this patient.

The long-term management of patients after TEVAR surgery should be given careful attention. Our patient developed graft infection 6 years after stent implantation, and the onset of infection was extremely insidious. By the time the etiology was definitively determined, the condition had become difficult to control. According to the guidelines for postendovascular repair imaging surveillance, imaging examinations should be conducted at 30 days, 6 months, and 12 months postsurgery (32). If no complications are found, imaging should be performed annually thereafter. In clinical practice, vigilance should be maintained for the possibility of complications when patients exhibit symptoms such as fever and pain after stent placement, and laboratory and imaging examinations should be promptly performed. For patients who have already developed complications, interdisciplinary consultations should be sought, and the most appropriate clinical treatment plan should be devised for the patient.

In summary, the therapeutic prospects of TEVAR for aortic diseases are promising, but this procedure also poses significant safety concerns. Enhancing the understanding of rare complications following TEVAR, monitoring the long-term outcomes of patients treated with TEVAR, and focusing on the prevention and timely diagnosis of post-TEVAR complications are of paramount importance.

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 humans were approved by the Ethics Committee of the 1st Hospital of Jilin University, China. The studies were conducted in accordance with the local legislation and institutional requirements. 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

DP: Validation, Writing – original draft. GX: Visualization, Writing – review & editing. BW: Validation, Writing – review & editing. XL: Supervision, Writing – review & editing. YW: Conceptualization, Project administration, Writing – review & editing.

Funding

The author(s) declare financial support was received for the research, authorship, and/or publication of this article. This work was supported by the National Natural Science Foundation of China [No. 81801972 to YW], Science and the Technology Research Project of Jilin Provincial Department of Education [JKH20211179KJ to YW], the Jilin Provincial Nature Science Foundation of Jilin Provincial Department of Science and Technology [20210101341JC to YW].

Acknowledgments

The authors are grateful to for the helpful discussions.

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.

Generative AI statement

The author(s) declare that no Generative AI was used in the creation of this manuscript.

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References

1. Riambau V, Böckler D, Brunkwall J, Cao P, Chiesa R, Coppi G, et al. Editor's choice - management of descending thoracic aorta diseases: clinical practice guidelines of the European society for vascular surgery (ESVS). *Eur J Vasc Endovasc Surg.* (2017) 53:4–52. doi: 10.1016/j.ejvs.2016.06.005

2. Upchurch GR Jr, Escobar GA, Azizzadeh A, Beck AW, Conrad MF, Matsumura JS, et al. Society for vascular surgery clinical practice guidelines of thoracic endovascular aortic repair for descending thoracic aortic aneurysms. *J Vasc Surg.* (2021) 73:55S–83. doi: 10.1016/j.jvs.2020.05.076

3. Trimarchi S, Gleason TG, Brinster DR, Bismuth J, Bossone E, Sundt TM, et al. Editor's choice - trends in management and outcomes of type B aortic dissection: a report from the international registry of aortic dissection. *Eur J Vasc Endovasc Surg.* (2023) 66:775–82. doi: 10.1016/j.ejvs.2023.05.015

4. Gopaldas RR, Huh J, Dao TK, LeMaire SA, Chu D, Bakaeen FG, et al. Superior nationwide outcomes of endovascular versus open repair for isolated descending thoracic aortic aneurysm in 11,669 patients. *J Thorac Cardiovasc Surg.* (2010) 140:1001–10. doi: 10.1016/j.jtcvs.2010.08.007

5. Schermerhorn ML, O'Malley AJ, Jhaveri A, Cotterill P, Pomposelli F, Landon BE. Endovascular vs. open repair of abdominal aortic aneurysms in the medicare population. *N Engl J Med.* (2008) 358:464–74. doi: 10.1056/NEJMoa0707348

6. Rey Chaves CE, Rojas S, Rosso JD, Peláez M, Sánchez EF, Hernández Rodríguez OG. Aortoesophageal fistulae following TEVAR: case report and literature review. *Int J Surg Case Rep.* (2023) 106:108126. doi: 10.1016/j.ijscr.2023.108126

7. Nakayama T, Nakamura Y, Yasumoto Y, Yoshiyama D, Kuroda M, Nishijima S, et al. Collapse of Najuta endograft (aortic arch graft) in patient with intermittent paraparesis. *Indian J Thorac Cardiovasc Surg.* (2022) 38:521–4. doi: 10.1007/s12055-022-01363-w

8. Karaolanis G, Moris D, Georgakarakos E, Donas K. Stroke after thoracic endovascular aortic aneurysm repair: a silent enemy that needs more caution. *Curr Pharm Des.* (2022) 28:798–9. doi: 10.2174/1381612828666220225121742

9. Faries PL, Cadot H, Agarwal G, Kent KC, Hollier LH, Marin ML. Management of endoleak after endovascular aneurysm repair: cuffs, coils, and conversion. *J Vasc Surg.* (2003) 37:1155–61. doi: 10.1016/S0741-5214(03)00084-3

10. Tzortzis E, Hinchliffe RJ, Hopkinson BR. Adjunctive procedures for the treatment of proximal type I endoleak: the role of peri-aortic ligatures and Palmaz stenting. *J Endovasc Ther.* (2003) 10:233–9. doi: 10.1177/152660280301000211

11. Tonnessen BH, Sternbergh WC 3rd, Money SR. Mid- and long-term device migration after endovascular abdominal aortic aneurysm repair: a comparison of AneuRx and Zenith endografts. *J Vasc Surg.* (2005) 42:392–400. doi: 10.1016/j.jvs.2005.05.040; discussion 400–1.

12. Deaton DH. Improving proximal fixation and seal with the HeliFx aortic EndoAnchor. *Semin Vasc Surg.* (2012) 25:187–92. doi: 10.1053/j.semvascsurg.2012.09.002

13. Jonker FH, Schlosser FJ, Geirsson A, Sumpio BE, Moll FL, Muhs BE. Endograft collapse after thoracic endovascular aortic repair. *J Endovasc Ther.* (2010) 17:725–34. doi: 10.1583/10-3130.1

14. Koda Y, Yamanaka K, Omura A, Gentsu T, Yamaguchi M, Okada K. Spinal cord ischemia after elective endovascular abdominal aortic aneurysm repair in a patient with multiple occlusions of the intercostal and internal iliac arteries. *J Vasc Surg Cases Innov Tech.* (2022) 8:447–9. doi: 10.1016/j.jvscit.2022.06.007

15. Dias-Neto M, Reis PV, Rolim D, Ramos JF, Teixeira JF, Sampaio S. Strategies to prevent TEVAR-related spinal cord ischemia. *Vascular.* (2017) 25:307–15. doi: 10.1177/1708538116671235

16. Spanos K, Kellert L, Rantner B, Banafsche R, Tsilimparis N. The importance of definitions and reporting standards for cerebrovascular events after thoracic endovascular aortic repair. *J Endovasc Ther.* (2018) 25:737–9. doi: 10.1177/1526602818808525

17. Daye D, Walker TG. Complications of endovascular aneurysm repair of the thoracic and abdominal aorta: evaluation and management. *Cardiovasc Diagn Ther.* (2018) 8:S138–56. doi: 10.21037/cdt.2017.09.17

18. Ibrahim A, Marchiori E, Eierhoff T, Martens S, Motekallemi A, Rukosujew A, et al. Post-implantation syndrome after frozen elephant trunk is associated with the

volume of new-onset aortic thrombus. *J Thorac Dis*. (2021) 13:4311–21. doi: 10.21037/jtd-21-398

19. Lyons OT, Patel AS, Saha P, Clough RE, Price N, Taylor PR. A 14-year experience with aortic endograft infection: management and results. *Eur J Vasc Endovasc Surg*. (2013) 46:306–13. doi: 10.1016/j.ejvs.2013.04.021

20. Sugiyama K, Iwahashi T, Koizumi N, Nishibe T, Fujiyoshi T, Ogino H. Surgical treatment for secondary aortoesophageal fistula. *J Cardiothorac Surg*. (2020) 15(1):251. doi: 10.1186/s13019-020-01293-x

21. Takeno S, Ishii H, Nanashima A, Nakamura K. Aortoesophageal fistula: review of trends in the last decade. *Surg Today*. (2020) 50:1551–9. doi: 10.1007/s00595-019-01937-z

22. Lyons OT, Baguneid M, Barwick TD, Bell RE, Foster N, Homer-Vanniasinkam S, et al. Diagnosis of aortic graft infection: a case definition by the management of aortic graft infection collaboration (MAGIC). *Eur J Vasc Endovasc Surg*. (2016) 52:758–63. doi: 10.1016/j.ejvs.2016.09.007

23. Chakfé N, Diener H, Lejay A, Assadian O, Berard X, Caillon J, et al. Editor's choice - European society for vascular surgery (ESVS) 2020 clinical practice guidelines on the management of vascular graft and endograft infections. *Eur J Vasc Endovasc Surg*. (2020) 59:339–84. doi: 10.1016/j.ejvs.2019.10.016

24. Laohapensang K, Arworn S, Orrapin S, Reanpang T, Orrapin S. Management of the infected aortic endograft. *Semin Vasc Surg*. (2017) 30:91–4. doi: 10.1053/j.semvascsurg.2017.11.001

25. Smeds MR, Duncan AA, Harlander-Locke MP, Lawrence PF, Lyden S, Fatima J, et al. Treatment and outcomes of aortic endograft infection. *J Vasc Surg*. (2016) 63:332–40. doi: 10.1016/j.jvs.2015.08.113

26. Chaufour X, Gaudric J, Goueffic Y, Khodja RH, Feugier P, Malikov S, et al. A multicenter experience with infected abdominal aortic endograft explantation. *J Vasc Surg*. (2017) 65:372–80. doi: 10.1016/j.jvs.2016.07.126

27. Kim YW. Aortic endograft infection: diagnosis and management. *Vasc Specialist Int*. (2023) 39:26. doi: 10.5758/vsi.230071

28. Laser A, Baker N, Rectenwald J, Eliason JL, Criado-Pallares E, Upchurch GR Jr. Graft infection after endovascular abdominal aortic aneurysm repair. *J Vasc Surg*. (2011) 54:58–63. doi: 10.1016/j.jvs.2010.11.111

29. Szeto WY, Desai ND, Moeller P, Moser GW, Woo EY, Fairman RM, et al. Reintervention for endograft failures after thoracic endovascular aortic repair. *J Thorac Cardiovasc Surg*. (2013) 145:S165–70. doi: 10.1016/j.jtcvs.2012.11.046

30. Kouijzer IJE, Baranelli CT, Maat I, van den Heuvel FMA, Aarntzen E, Smith T, et al. Thoracic aortic vascular graft infection: outcome after conservative treatment without graft removal. *Eur J Cardiothorac Surg*. (2022) 63(1):ezac551. doi: 10.1093/ejcts/ezac551

31. Roy D, Grove DI. Efficacy of long-term antibiotic suppressive therapy in proven or suspected infected abdominal aortic grafts. *J Infect*. (2000) 40:184–7. doi: 10.1016/S0163-4453(00)80014-6

32. 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: executive summary. 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. *Catheter Cardiovasc Interv*. (2010) 76:E43–86. doi: 10.1002/ccd.22537

frontiers | Frontiers in Cardiovascular Medicine

TYPE Case Report
PUBLISHED 28 February 2025
DOI 10.3389/fcvm.2025.1498359

OPEN ACCESS

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RECEIVED 18 September 2024
ACCEPTED 07 February 2025
PUBLISHED 28 February 2025

CITATION
Shan J, Wang H and Xue S (2025) Case Report: "Dumbbell" giant right coronary artery ectasia with right atrial fistula.
Front. Cardiovasc. Med. 12:1498359.
doi: 10.3389/fcvm.2025.1498359

Case Report: "Dumbbell" giant right coronary artery ectasia with right atrial fistula

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A 50-year-old female patient presented with a "dumbbell" giant right coronary artery ectasia, characterized by two artery dilation segments both reaching the level of a giant aneurysm with a normal segment between them. Computed tomography angiography showed a fistula sac in the right atrium. The vessel shape was a typical type IV (localized or segmental) coronary artery ectasia, which is rarely seen on true imaging. The patient had a 3-year history of chest tightness, without dyspnea, worsened by physical activity. Additional tests indicated that she had mitral valve regurgitation, superficial myocardial bridge, and anemia, all of which led to the development of her symptoms. She felt relieved after successful coronary artery fistula repair, mitral valvuloplasty, and fistula sac removal. At the 6-month follow-up, no complications were found according to echocardiography. Patients with coronary aneurysms can be asymptomatic in the early stage, while this case indicates that the dumbbell shape may be a developing stage of giant coronary aneurysm whose origin and close-fistula segments are influenced by separate hydrodynamics during ectasia or aneurysm formation.

KEYWORDS

cardiovascular surgery, case report, computed tomography angiography, coronary artery ectasia, coronary artery fistula

1 Introduction

Coronary artery fistula has an estimated prevalence of 0.002% in the general population, and fistula drainage sites can be found in the aorta, pulmonary artery, and all four heart chambers (1). Coronary artery ectasia, defined as diffuse vessel dilation >1.5 times that of the adjacent normal segment, is a specific coronary aneurysmal dilation with a prevalence in the range of 1.2%–4.9% (2). Some patients can be asymptomatic with coronary artery ectasia and/or fistula, while surgical intervention is necessary and can be effective in cases with cardiovascular comorbidities to prevent further complications.

Here, our team reports a rare case of type IV segmental ectasia with dilating segments located at both ends of the right coronary artery (RCA), from the origin to the fistula opening, which lacks sufficient real images (3). Both dilating segments met the criteria for giant coronary artery aneurysm [internal diameter (ID) >20 mm or >4× ref. ID] (3).

2 Case presentation

2.1 Patient information and timeline

A 50-year-old woman had a 3-year history of chest tightness without dyspnea. Her symptoms worsened after physical activity. She denied any past medical history of hypertension or diabetes. No significant family or psychosocial history was reported. She was menopausal on arrival at the hospital.

The patient reported having an echocardiogram 3 years before at a local institution, which indicated an RCA fistula to the right atrium (RA) and mitral regurgitation; however, no therapy was provided as she was unwilling to undergo a thoracotomy at that time. As her symptoms worsened, she had a second echocardiogram with similar results at another local institution 1 month before this admission, and she decided to receive treatment. She was initially admitted to the cardiology department of our institution and then transferred to our department 3 days later.

2.2 Clinical findings and diagnostic assessment

The patient's vital signs upon admission were as follows: axillary temperature 36.4°C; heart rate 76 beats/min; respiratory rate 17 breaths/min; and blood pressure 98/43 mmHg. Transthoracic echocardiography (TTE) showed a left ventricular ejection fraction (LVEF) of 67% (ref. ≥54%). Laboratory tests showed a brain natriuretic peptide (BNP) level of 111.0 pg/mL (ref. 0.0–100.0 pg/mL), a platelet count of 397×10^9/L (ref. 125–350×10^9/L), and a lipoprotein (a) level of 892 mg/L (ref. 0.0–300.0 mg/L).

Computed tomography angiography and digital subtraction angiography showed a "dumbbell" RCA ectasia at the origin (ID 14 mm) and the fistula wall in the RA (ID 37mm × 36 mm, forming a fistula sac), and the connecting segment was normal (Figure 1, Supplementary Material Video S1). The ostium from the RCA fistula sac to the RA was 10 mm in width, facing the inferior vena cava. Transesophageal echocardiogram (TEE) showed a continuous flow enhanced during diastole at the entrance (peak velocity 2.5 m/s) and exit (peak velocity 1.8 m/s) of the RCA fistula sac (Figure 2, Supplementary Material Videos S2, S3). Her atria (left ID 44 mm, ref. 27–38 mm; right vertical diameter 62 mm, ref. <53 mm), left ventricle (end of diastole ID 61 mm, ref. 39–52 mm), and pulmonary artery (ID 32 mm, ref. 12–26 mm) were enlarged.

Additional laboratory and auxiliary tests indicated the following: (1) anemia (hemoglobin 71 g/L), with lowered mean corpuscular volume (MCV) of 66.5 fl (ref. 82–100 fl), mean corpuscular hemoglobin (MCH) of 17.4 pg (ref. 27–34 pg), mean corpuscular hemoglobin concentration (MCHC) of 264 g/L (ref. 316–354 g/L), and lowered ferritin of 5.4 μg/L (ref. 11–306.8 μg/L), serum iron of 2.5 μmol/L (ref. 7.8–32.2 μmol/L); (2) moderate mitral regurgitation (with an enlarged mitral annulus of 38 mm, ref. 21–32 mm); and (3) superficial myocardial bridge in the left anterior descending branch [50% compression during systole, thrombolysis in myocardial infarction (TIMI) flow grade III]. These cardiovascular comorbidities may have complicated effects on symptom development. No evidence of coronary atherosclerosis, infective endocarditis, or pulmonary disease was found.

2.3 Therapeutic intervention and follow-up

The patient received a dual antiplatelet therapy (100 mg aspirin, 75 mg clopidogrel, q.d., PO), atorvastatin calcium tablets (20 mg/tablet, q.n., PO), rabeprazole sodium enteric-coated capsules (10 mg/capsule, q.d., PO), potassium chloride sustained release tablets (0.5 g/tablet × 2, b.i.d., PO), ferrous succinate tablets (0.4 g/tablet, q.d., PO) for the first 3 days. After being transferred to our department, the patient received recombinant human erythropoietin (10^4 IU, q.o.d., SC); ferrous succinate tablets (0.2 g/tablet, q.d., PO) were also given for anemia. The hemoglobin (76 g/L), MCV (64.7 fl), MCH (17.2 pg), and MCHC (266 g/L) were not much improved 1 day before surgery.

Five days after transfer, the patient underwent cardiopulmonary bypass-assisted coronary artery fistula repair and mitral valvuloplasty (Figure 3), and the RA fistula sac was removed. Her postoperative TEE showed no RA fistula flow (Supplementary Videos S4, S5). The patient experienced symptom relief postoperatively. Since anemia may increase the risk of postoperative cardiovascular events, acute kidney injury, and mortality, indices including D-dimer, creatinine, and BNP were monitored (4). Her anemia did not significantly worsen postoperatively (hemoglobin 71 g/L). After a 10-day hospital stay, she was discharged with prescriptions for warfarin sodium tablets (2.5 mg/tablet, q.d., PO), digoxin tablets (0.13 mg, q.d., PO), metoprolol tartrate tablets (12.5 mg, b.i.d., PO), spironolactone tablets (20 mg/tablet, b.i.d., PO), potassium chloride sustained release tablets (0.5 g/tablet × 2, b.i.d., PO), and hydrochlorothiazide tablets (25 mg/tablet × 2, q.d., PO).

Six months after discharge, the patient experienced no cardiovascular symptoms. She self-reported having no complications or other health events. The TTE found no regurgitation or other complications at the mitral valve, and the LVEF was 69%. The fistula was well repaired with no leakage, and the ID of the RCA origin remained at 15 mm. The patient chose not to have additional invasive examinations.

3 Discussion

The RCA is involved in 50%–60% of coronary artery fistulae, and 19%–26% of fistula drainage sites are located in the RA (1). The RCA-RA fistula can lead to aneurysm formation. Of patients with coronary artery fistula, 5.9% experience a coronary artery aneurysm (5). A computer simulation of hydrodynamics in an

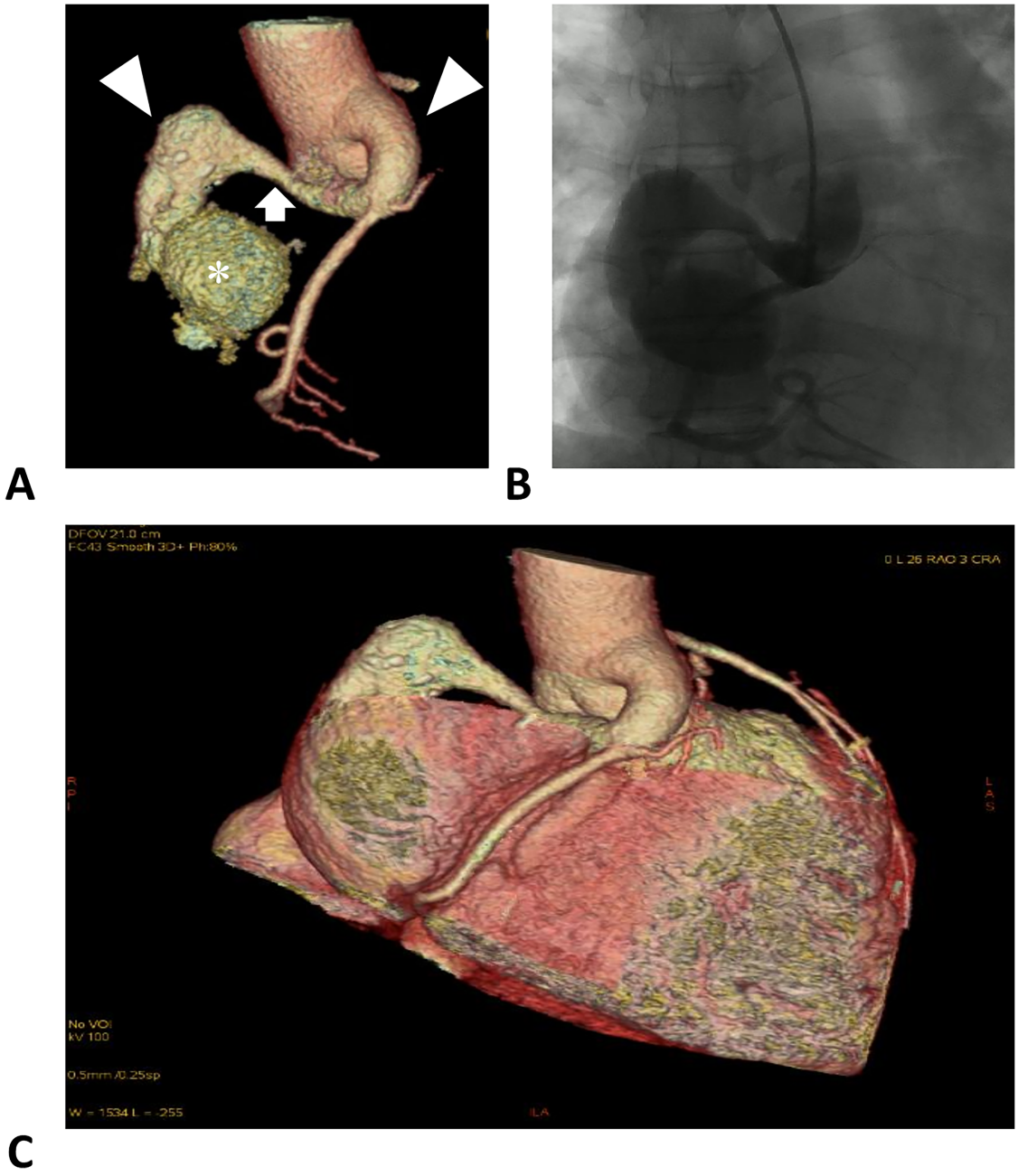

FIGURE 1
Right coronary artery images. **(A)** Computed tomographic angiography of the RCA. The left and right triangles denote the distal and proximal aneurysmal segments of the RCA. The arrow denotes a normal segment between the dilations, forming a dumbbell-shaped ectasia. The asterisk denotes the fistula sac formed in the right atrium. **(B)** Digital subtraction angiography of the RCA. **(C)** Reconstruction of the coronary arteries and the heart, illustrating the positional relationship.

RCA-right ventricle fistula model found (1) high fluid velocity and large wall shear stress, and (2) high pressure at the distal and proximal RCA segments, respectively, and both abnormalities can be corrected by fistula closure (6). Except for this specific case, the initial anatomy and local vulnerability of coronary arteries may affect dilation development, forming different aneurysm shapes (7). The dumbbell-shaped dilation of the RCA found in our patient may have been influenced by its unique hydrodynamics. Other adult congenital cases of RCA fistula presented similar RCA deformation (Supplementary Table S1). Interestingly, RCA origin or proximal segment dilation and distal giant aneurysm can exist separately or simultaneously, indicating that they may be influenced by independent fluid forces. It is reasonable to assume that, without correction, the hydrodynamic aberration could have continued to accelerate the progression of dilation.

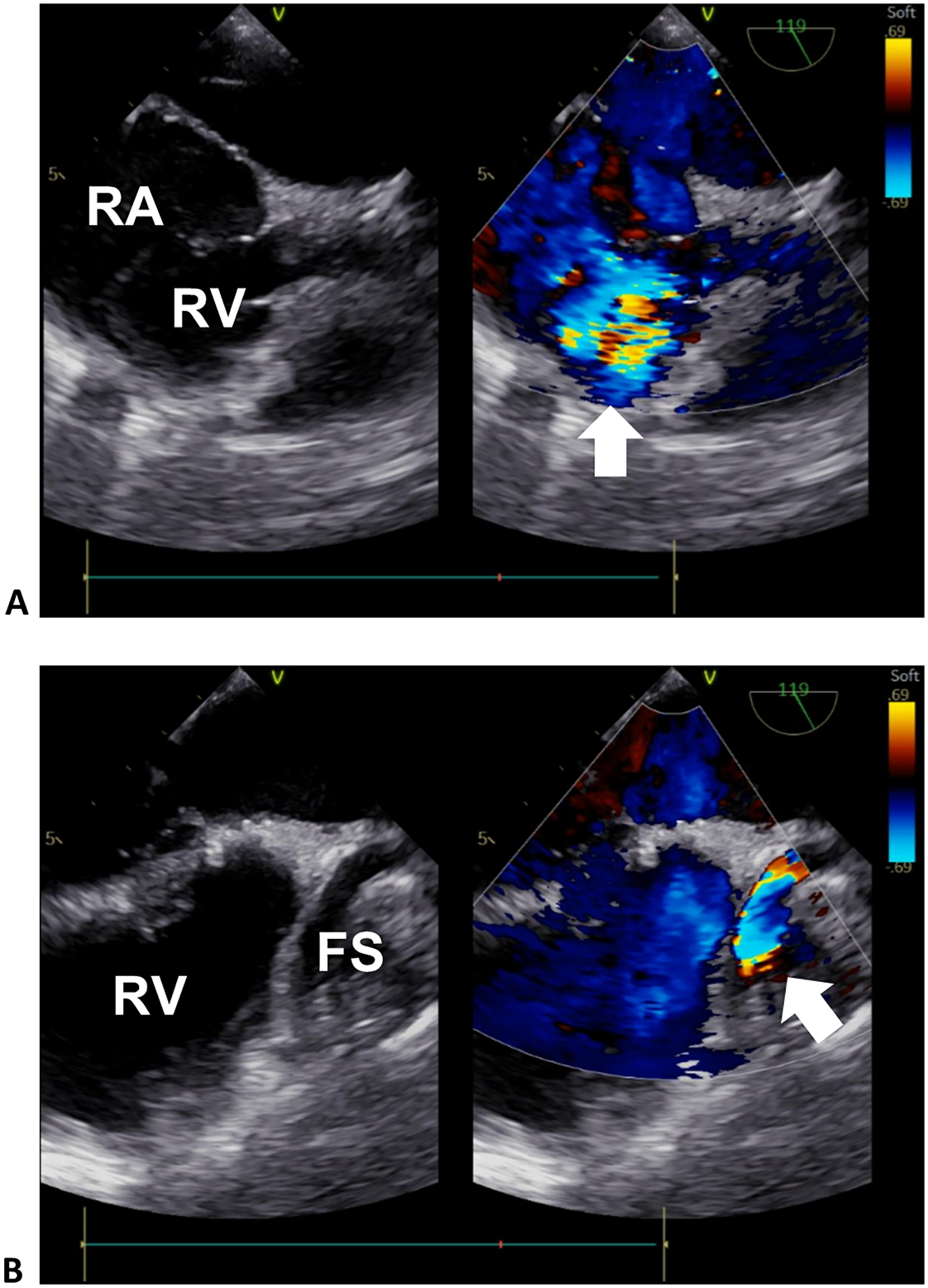

FIGURE 2
Transesophageal echocardiogram of the right heart. **(A)** Sectional plane showing the exit flow from the fistula sac into the right heart. **(B)** Sectional plane showing the entrance flow from the RCA to the fistula sac. The arrows denote the turbulence signals of the exit and entrance flows in **(A)** and **(B)**, respectively. RA, right atrium; RV, right ventricle; FS, fistula sac.

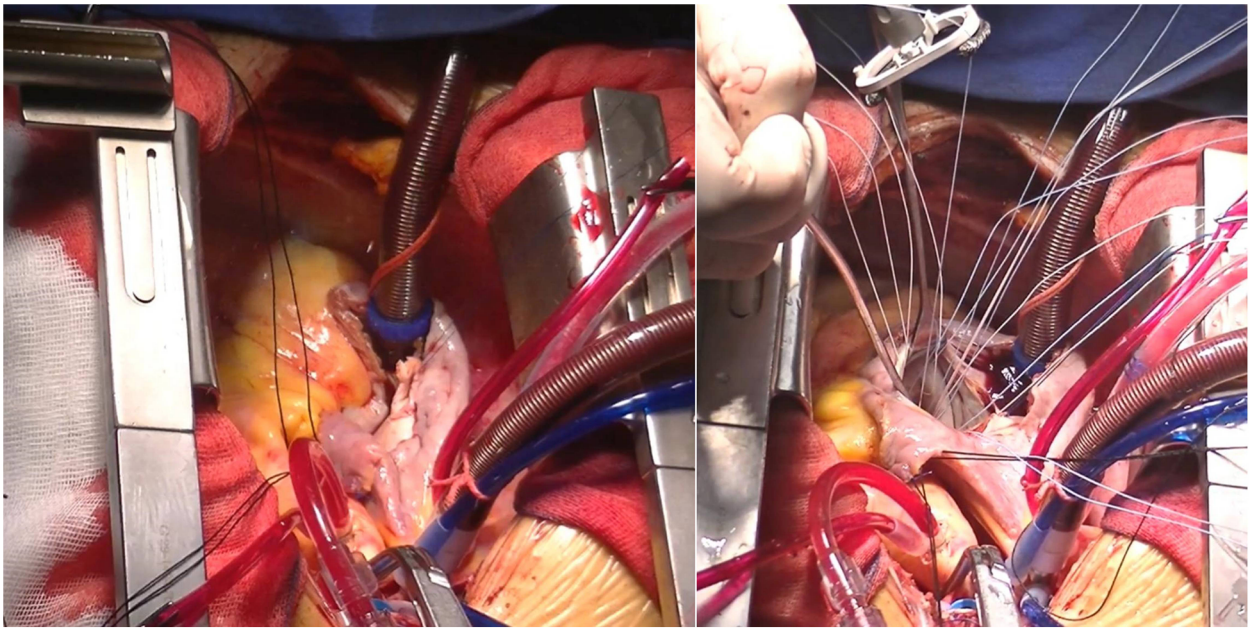

FIGURE 3
Surgical procedures. Left panel: right coronary artery to right atrial fistula repair. Right panel: mitral valvuloplasty.

A number of patients with coronary artery fistula can be asymptomatic, especially in the early stages of giant aneurysm formation. However, several factors can lead to significant symptoms or signs. First, the turbulent flow can result in local thrombogenesis, potentially leading to acute coronary syndrome (8, 9). Second, the enlarged RCA can exert an occupying effect that compresses adjacent tissues. Third, blood diversion can cause myocardial ischemia, functional valve regurgitation, infective endocarditis, and late heart chamber enlargement, which increases the risk of heart failure and arrhythmia (10). Dyspnea and chest pain are the most common symptoms reported in RCA fistula cases, while palpitations, fever, and other infectious symptoms can also be seen in these patients (Supplementary Table S1). Although functional tricuspid or pulmonary valve regurgitation is more common in RCA-RA fistula for the left-to-right diversion, our case had an uncommon mitral valve regurgitation instead. Similar to a previous report of a patient with an early-stage RCA-RA fistula and relatively normal morphology but mitral insufficiency who experienced chest pain, the mitral valve regurgitation in our patient may have contributed to her chest tightness (11). In addition, the myocardial bridge can add to myocardial ischemia caused by blood diversion, which exacerbates the symptom during exertion. Our team concluded that the patient's symptoms resulted from the combined effects of multiple heart conditions.

In conclusion, the dumbbell shape may represent an early stage in the development of a larger giant RCA aneurysm, with the RCA origin and close-fistula segments being influenced by separate hydrodynamic forces during ectasia formation.

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 humans were approved by the Ethics Committee of Renji Hospital, School of Medicine, Shanghai Jiao Tong University. The studies were conducted in accordance with the local legislation and institutional requirements. The 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

JS: Conceptualization, Data curation, Writing – original draft. HW: Formal Analysis, Investigation, Writing – original draft. SX: Resources, Writing – review & editing, Supervision.

Funding

The author(s) declare that no financial support was received for the research, authorship, and/or publication of this article.

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.2025.1498359/full#supplementary-material

References

1. 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

2. Devabhaktuni S, Mercedes A, Diep J, Ahsan C. Coronary artery ectasia-a review of current literature. *Curr Cardiol Rev.* (2016) 12:318–23. doi: 10.2174/1573403X12666160504100159

3. Kawsara A, Núñez Gil I, Alqahtani F, Moreland J, Rihal C, Alkhouli M. Management of coronary artery aneurysms. *JACC Cardiovasc Interv.* (2018) 11:1211–23. doi: 10.1016/j.jcin.2018.02.041

4. Kloeser R, Buser A, Bolliger D. Treatment strategies in anemic patients before cardiac surgery. *J Cardiothorac Vasc Anesth.* (2023) 37:266–75. doi: 10.1053/j.jvca.2022.09.085

5. Li D, Wu Q, Sun L, Song Y, Wang W, Pan S, et al. Surgical treatment of giant coronary artery aneurysm. *J Thorac Cardiovasc Surg.* (2005) 130:817–21. doi: 10.1016/j.jtcvs.2005.04.004

6. Cao H, Qiu Y, Yuan D, Yu J, Li D, Jiang Y, et al. A computational fluid dynamics study pre- and post-fistula closure in a coronary artery fistula. *Comput Methods Biomech Biomed Engin.* (2020) 23:33–42. doi: 10.1080/10255842.2019.1699540

7. Rafiei A, Saidi M. Aneurysm geometric features effect on the hemodynamic characteristics of blood flow in coronary artery: CFD simulation on ct angiography-based model. *Med Biol Eng Comput.* (2022) 60:3357–75. doi: 10.1007/s11517-022-02676-z

8. Khalil G, Elbadri A, Abbasi S, Das I, Ladwiniec A. An unusual cause of acute coronary syndrome: thrombosis of right coronary artery to right atrium fistula. *BMJ Case Rep.* (2023) 16:E257368. doi: 10.1136/bcr-2023-257368

9. Shirai K, Ogawa M, Kawaguchi H, Kawano T, Nakashima Y, Arakawa K. Acute myocardial infarction due to thrombus formation in congenital coronary artery fistula. *Eur Heart J.* (1994) 15:577–9. doi: 10.1093/oxfordjournals.eurheartj.a060546

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

11. Koukash M, Hamad M, Makhlouf A, Kakaje A, Alsaid B. Double right coronary artery with fistula to the right atrium and mitral insufficiency: a case report. *Int J Surg Case Rep.* (2022) 96:107386. doi: 10.1016/j.ijscr.2022.107386

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TYPE Case Report
PUBLISHED 28 April 2025
DOI 10.3389/fcvm.2025.1530548

OPEN ACCESS

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RECEIVED 19 November 2024
ACCEPTED 08 April 2025
PUBLISHED 28 April 2025

CITATION
Liang C, Liu H, Cao Z, Li T, Wang Y, Xiao Y and Ma R (2025) Successful treatment of massive biventricular thrombi associated with myocarditis: a case report.
Front. Cardiovasc. Med. 12:1530548.
doi: 10.3389/fcvm.2025.1530548

Successful treatment of massive biventricular thrombi associated with myocarditis: a case report

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Background: Massive biventricular thrombi associated with myocarditis are rare and pose significant management challenges.
Case presentation: A 24-year-old male from a plateau region presented with dyspnea, chest pain, and cerebral infarction/hemorrhage. Imaging revealed giant biventricular thrombi (left: 81 × 62 × 59 mm; right: 59 × 16 mm) and LVEF of 25%.
Management: Multidisciplinary therapy included anticoagulation and GDMT.
Outcomes: Complete thrombus resolution occurred by 10 weeks, with improved cardiac function (LVEF 48%) and no recurrent embolism.
Conclusion: Drug therapy may be effective for massive biventricular thrombi in myocarditis.

KEYWORDS
biventricular thrombi, myocarditis, cerebral infarction, cerebral hemorrhage, anticoagulation

Introduction

Ventricular thrombi are rare but carry a high risk of systemic embolism and mortality (1). Myocarditis, characterized by inflammatory infiltration of the myocardium, may lead to endothelial injury, blood stasis due to impaired ventricular function, and a hypercoagulable state—factors that align with Virchow's triad and predispose patients to thrombus formation (2). The resolution of small mural (laminated) ventricular thrombus is predominantly accomplished through anticoagulation (2), however, the management of large ventricular thrombus remains uncertain, with only a limited number of cases documented in the literature (3–11). This case represents the largest reported instance of biventricular thrombi, complicated by cerebral infarction and hemorrhage, and the treatment process was notably complex. This case aims to demonstrate the feasibility of medical therapy for massive biventricular thrombi in myocarditis, particularly in patients with concurrent cerebral infarction and hemorrhage, and to highlight the challenges in balancing anticoagulation risks.

Case presentation

A 24-year-old male resident of the plateau region presented to the emergency department with a 2-day history of weakness and anorexia, following a month-long period of dyspnea, chest pain, and dry cough. The patient was conscious and exhibited stable vital signs, including a temperature of 36.6°C, a pulse rate of 100 bpm, a

respiratory rate of 18 bpm, and a blood pressure of 152/84 mmHg. The physical examination revealed an arrhythmia (ventricular premature beats) without associated murmurs, and the presence of lower limb edema, but no jugular vein distension. The patient had a history of smoking 1 pack a day for 4 years and ceased smoking 7 months prior.

Laboratory test revealed a hemoglobin of 187 g/L, a hematocrit of 56.4%, a erythrocytes count number of 6.67×10^{12}/L, a positive troponin assay, a C-reactive protein (CRP) level of 68.62 mg/L, a procalcitonin of 0.34 ng/ml, an albumin level of 22.3 g/L, and a B-type natriuretic peptide (BNP) of 1,370 pg/ml. Thromboelastography, coagulation tests, tumor markers test, immune-related test (including plasma protein S, plasma protein C, anticoagulant antibody spectrum, anti-cardiolipin antibody, anti-streptolysin O) were within normal. Thrombophilia tests were not performed due to conditions being limited. A Electrocardiograph indicated ST segment changes and ventricular premature beats. A transthoracic echocardiography (TTE) revealed a anteroposterior diameter of 62 mm for the left ventricle and 55 mm for the right ventricle. The examination revealed that the tricuspid valve regurgitation was moderate, with a regurgitation velocity of 237 cm/s and a peak pressure gradient of 22 mmHg. The left ventricle ejection fraction (LVEF) was measured at 25%. The tricuspid annular plane systolic excursion (TAPSE) was measured to be 12 mm. Additionally, the presence of a potential biventricular thrombi was detected (left ventricular thrombus dimensions: 81 mm × 62 mm × 59 mm; right ventricular thrombus dimensions: 59 mm × 16 mm) (Figure 1a). Cardiac computed tomography angiography (CTA) revealed a low-density shadow indicative of a biventricular mass (Figure 1b), raising the possibility of thrombus. No abnormalities were detected in the coronary arteries, pulmonary artery, or thoracic aorta. Additionally, severe pathology was noted in both lungs, accompanied by a small amount of fluid in the bilateral pleural cavities. 3D models are used to better understand ventricular thrombi (Figure 2). Positron Emission Tomography-Computed Tomography (PET-CT) scan revealed low-density shadows in both the left and right ventricles, with no abnormal increase in FDG metabolism. Multiple acute infarction were observed in the bilateral cerebellar hemispheres, alongside slightly high-density shadows indicative of infarction and hemorrhage. Pulmonary plaque shadows were noted, accompanied by increased FDG metabolism suggestive of inflammation. Additionally, diffuse pericardial FDG metabolism was observed, pointing to inflammatory lesions. Multiple mucosal effusions and systemic subcutaneous edema were also considered in the findings. The magnetic resonance imaging (MRI) myocardial perfusion imaging revealed the left and right ventricular filling defects, the potential presence of thrombus, the left ventricular wall and subendocardial late gadolinium enhancement (LGE), the possibility of myocarditis or cardiomyopathy, the reduced systolic function of the left ventricle with an LVEF of 17%, and the presence of a small

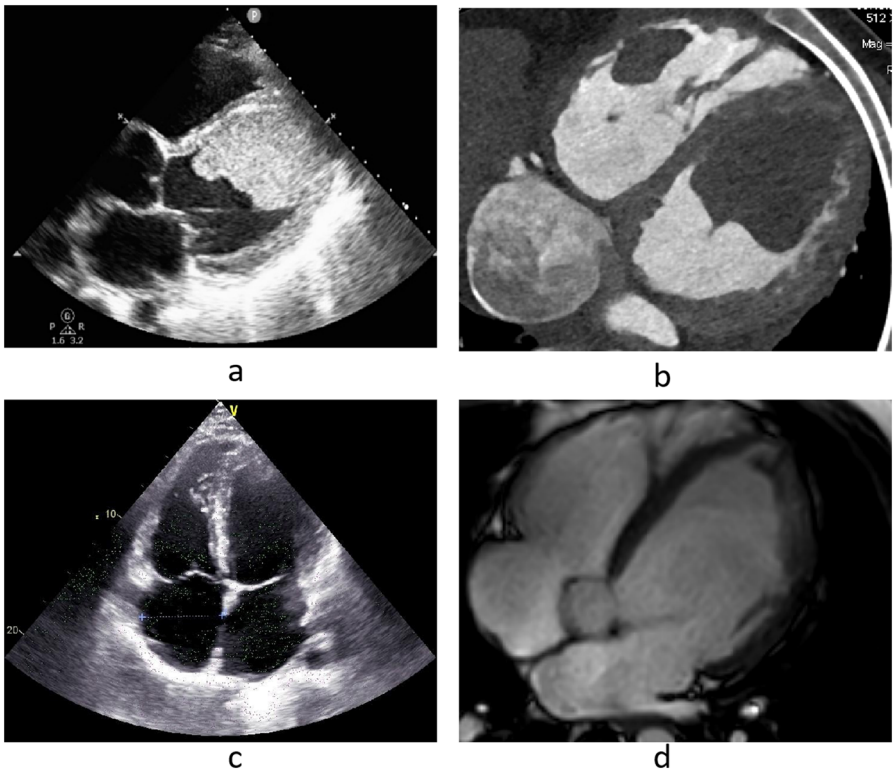

FIGURE 1
(a,b) Echocardiography and cardiac CTA on admission, (c,d) echocardiography and cardiac MRI before discharge.

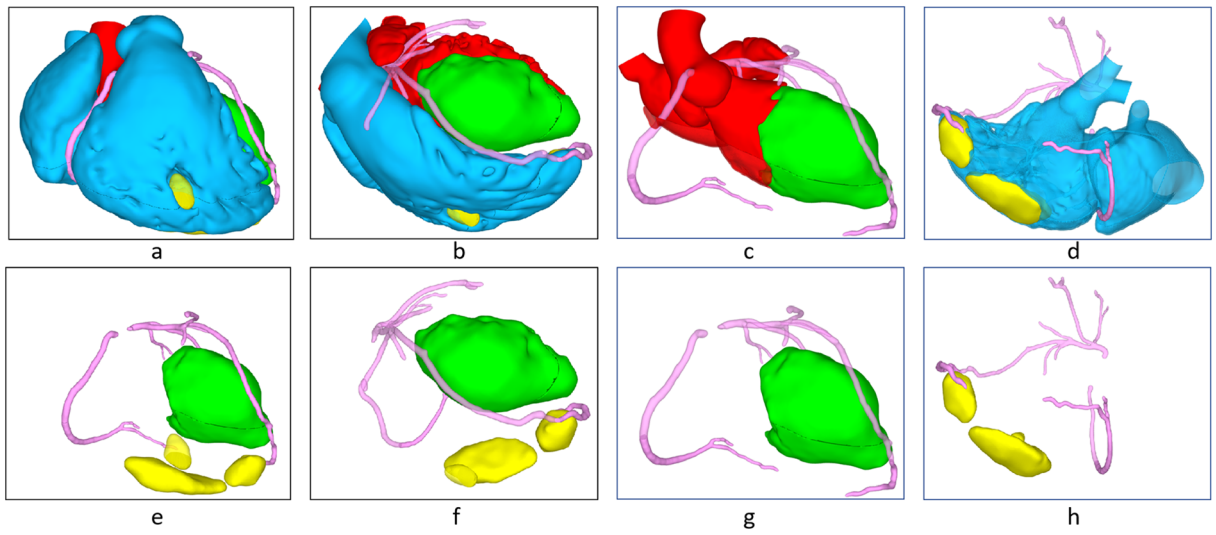

FIGURE 2
3D reconstructed image (red: left blood flow, blue: right blood flow, green: left ventricular thrombosis, yellow: right ventricular thrombosis), (a,e) frontal view, (b,f) bertical interventricular septal plane, (c,g) left ventricualr outflow tract plane (d,h) right ventricular outflow tract plane.

amount of fluid in the pericardial and pleural cavities. Neuroimaging, including brain computed tomography (CT) and diffusion-weighted imaging (DWI), revealed multiple acute cerebral infarctions in the bilateral fronto-parietal and occipital lobes, the left thalamus, and the bilateral cerebellum, along with a small amount of hemorrhage in the right cerebellum. Cervical vascular ultrasound revealed the presence of mural thrombus in the right internal jugular vein catheter, measuring approximately 0.6 cm × 2.8 cm.

Treatment strategy

Following a multidisciplinary consultation, drug therapy was recommended, however, patients may experience systemic embolism and pulmonary embolism in the course of treatment. While the patient was administered a low a dose of enoxaparin (40 mg BID) due to the presence of cerebral hemorrhage, metoprolol 23.75 mg QD, spironolactone 20 mg QD, dapagliflozin 10 mg QD, sacubitril/valsartan 24/26 mg BID for GDMT, along with other supportive treatments. The patient underwent daily monitoring through TTE. After a 1-week treatment period, TTE revealed an increase in LVEF to 33% and in TAPSE to 14 mm. The size of the biventricular thrombi remained unchanged. Cervical vascular ultrasonography revealed the presence of thrombus not only in the right internal jugular vein but also in the left subclavian vein and the proximal segment of the left internal jugular vein, with significant mobility observed. Following a multidisciplinary discussion, considering that the risk of cerebral hemorrhage has decreased, but the risk of thrombosis is still very high, the therapeutic dosage of enoxaparin was adjusted to 70 mg (1 mg/kg) Q12H. CRP and procalcitonin returned to normal 10 days after admission.

Two weeks post-admission, TTE revealed a reduction in the left ventricular thrombus to 62 mm × 60 mm × 41 mm and the right ventricular thrombus to 46 mm × 18 mm. Additionally, the LVEF measured of 36%, and TAPSE was at 15 mm. However, there was no alteration in ventricular size, and mobile was noted on the free surface of the left ventricular thrombus. Cardiac CT revealed the absence of pulmonary embolism, while brain CT revealed a reduction in the cerebral infarction and no evidence of new hemorrhage. The patient exhibited no new signs or symptoms. Considering the patient's history of cerebral hemorrhage extending beyond 2 weeks, the risk associated with extracorporeal circulation surgery was significantly diminished. A multidisciplinary consensus recommended prioritizing surgical thrombectomy due to the observed mobility of left ventricular thrombus. The complexity of thrombectomy procedures results in extended operation times. The prolonged duration of the surgery can further compromise cardiac function, markedly elevating the risk of postoperative low cardiac output syndrome and cerebral hemorrhage. Consequently, the likelihood of requiring postoperative intra-aortic balloon pumping (IABP) or extracorporeal membrane oxygenation (ECMO) is significantly increased, which in turn raises the risk of cerebrovascular events.

Two days subsequent to the decision to perform a thrombectomy, TTE revealed a reduction in the thrombus size within the left ventricle to 56 mm × 45 mm × 38 mm and within the right ventricle to 19 mm × 13 mm. Additionally, cervical vascular ultrasound revealed a decrease in the thrombus size within the right internal jugular vein. No thrombus was detected in the left subclavian vein or the left internal jugular vein. The patient exhibited no new clinical symptoms. Following a multidisciplinary consultation, it was determined that the thrombus had significantly decreased in size, thereby permitting the continuation of medication and a transition from enoxaparin

to rivaroxaban for anticoagulation therapy. Nevertheless, the risk of embolization persisted.

Following sustained drug management, including anticoagulation and GDMT, cervical vascular ultrasound revealed complete resolution of the right internal jugular vein thrombus 3 weeks post-admission. Additionally, cardiac MRI revealed full resolution of the right ventricular thrombus at 4 weeks and the left ventricular thrombus at 10 weeks post-admission (Figure 1d). Brain CT revealed that the cerebral infarction reduced in size, with no indications of new cerebral infarctions or cerebral hemorrhages. Prior to discharge, TTE revealed normal size and function of the right ventricle, with a TAPSE of 20 mm. The anteroposterior diameter of the left ventricle measured at 68 mm, the LVEF was 48%, and no valvular regurgitation was detected, with a tricuspid regurgitation velocity of 346 cm/s and a peak pressure gradient of 48 mmHg. The patient did not present with any new symptoms of embolism, and there was a marked improvement in symptoms such as weakness, anorexia, dyspnea, chest pain and dry cough.

In the subsequent treatment regimen, the patient's dose of sacubitril/valsartan was adjusted to 36/39 mg BID, with no other drug modifications. The patient's vital signs included a resting heart rate of 70 BPM and a blood pressure of 100/70 mmHg. Given the patient's medical history of biventricular thrombus and myocarditis, it was recommended that the patient should avoid returning to work at high altitudes and continue rivaroxaban therapy for a duration of 6 months. In this cases of left heart-associated pulmonary hypertension, initial observation was conducted, and drug of pulmonary hypertension treatment was determined based on the results of subsequent patient evaluations.

Follow-up

The patients were followed up for 2 weeks, 1 month, 3 month and 6 month, and no new clinical symptoms appeared, resting heart rate was 65 BPM, blood pressure was 101/65 mmHg. TTE revealed the LVEF was 44%, and no valvular regurgitation was detected, with a tricuspid regurgitation velocity of 251 cm/s and a peak pressure gradient of 25 mmHg. Cardiac MRI showed no ventricular thrombus. The patient returned to daily life, but did not perform physical work.

Discussion

According to Virchow's triad, the pathogenesis of left ventricular thrombus involves the interplay of three critical factors: (1) blood stasis resulting from diminished ventricular function, (2) endocardial injury, (3) a state of inflammation or hypercoagulability. The relative contributions of these factors to the formation of LV thrombus depend on the etiology of myocardial dysfunction and its duration. While regional endocardial injury and inflammation may play a predominant role in an acute myocardial infarction (MI), stasis resulting from diminished LV function is likely to be the principal factor in dilated cardiomyopathy (DCM) (2). The patient in this case was considered myocarditis: did not have instrumental lesions, obvious clinical symptoms, troponin was positive, ST segment changed in ECG, and serious diminished in ventricular systolic function. He experienced persistent dyspnea and pain over a 1-month period, suggesting the presence of heart failure during that time. Despite the patient's young age and good physical condition, which allowed for tolerance of prolonged impaired ventricular systolic function, the myocarditis had also resulted in damage to the endocardial membrane. The risk of hypercoagulability is heightened in patients residing at high altitudes due to elevated hemoglobin levels. Three factors facilitate the development of biventricular thrombi in such patients. The adhesion of thrombus to the ventricular wall further impairs ventricular muscle contraction, exacerbating symptoms of nausea and weakness. This clinical presentation is indicative of recent thrombus formation. LV thrombus is reportedly less prevalent in DCM compared to ischemic cardiomyopathy. This discrepancy may be attributed to underdetection, as the incidence rates of thromboembolic events have been observed to be comparable between the two conditions (12). Limited data also suggest that protuberant thrombi may resolve earlier than mural thrombi [OR, 3.2 (95% CI, 1.1–8.89); $P = 0.026$] (13), which may affect long-term thromboembolic potential.

In our case, the complete dissolution of the right ventricular thrombus was observed within 4 weeks, and the left ventricular thrombus required 6 weeks. At present, giant ventricular thrombus is predominantly documented through case reports (3–11), with treatment strategies including anticoagulation, fibrinolysis, and surgical thrombectomy. Nevertheless, the lack of substantial evidence-based medical data has resulted in persistent debates concerning the most effective therapeutic approach (2). For patients demonstrating adequate cardiac function, surgical thrombectomy is typically recommended (9, 10). In instances of ischemic cardiomyopathy, where cardiac function is compromised, a combination of bypass surgery and thrombectomy is regarded as an efficacious treatment strategy (3). Conversely, for patients with severely impaired cardiac function, thrombolysis is the preferred option among most clinicians (4–8, 11). It is imperative to evaluate the risk of thrombus embolization and to make decisions through multidisciplinary discussions. In addition to ventricular thrombus concomitant with myocarditis, our patient presented with both cerebral infarction and hemorrhage, cerebral infarction is most likely caused by the loss of ventricular thrombus, followed by cerebral hemorrhage after infarction. Thereby markedly complicating the management of anticoagulation therapy and surgical intervention. Balancing the benefits and risks of these therapeutic approaches proved to be exceedingly challenging and complex. When low doses of enoxaparin were given in the early stage to prevent cerebral hemorrhage, the patient's ventricular thrombus did not change, and further venous thrombus occurred. After multidisciplinary discussion, the biventricular thrombi was gradually dissolved after adjusting the enoxaparin dose to normal. However, partial dissolution of ventricular

thrombus also leads to thrombus mobile, while GDMT treatment gradually restores the contractility of the left ventricle, which greatly increases the risk of ventricular thrombus drop out. Therefore, it is necessary to closely observe the symptoms of patients and timely deal with. If fatal embolism occurs, the consequences are unimaginable. However, performing a thrombectomy presents significant challenges. Firstly, achieving adequate exposure is difficult. While removal of the mitral valve can increase the visual field, it increase the risk of damaging mitral valve. Secondly, the thrombus in trabeculae carneae of the ventricles can't be completely removed, leaving residual thrombi could subsequently enter the systemic or pulmonary circulation once ventricular contractions resume. Lastly, the thrombus may strongly adherence to the endocardium, leading to residual roughened areas following the procedure. This condition further predisposes the patient to thrombosis. So it's a very difficult choice whether or not to surgical thrombectomy.

In the absence of extensive literature and guidelines, each step we undertake is approached with caution and complexity. Although we have successfully used rivaroxaban to dissolve ventricular thrombus, many clinicians are concerned that rivaroxaban cannot be routinely monitored to assess whether therapeutic effect has been achieved, especially in places with poor medical conditions. Same with other non-vitamin-K-antagonist oral anticoagulants. For the current literature on giant biventricular thrombus, all of them are successful in thrombolysis, and there are few data on serious complications or failures in the treatment process, which seriously affects the formulation of current guidelines and consensus. In addition, thrombus and ventricular contractility interact with each other, so whether myocardial contractility can be further improved after the removal of thrombus, the surgical thrombectomy is not getting more support.

Conclusion

This case demonstrates that aggressive anticoagulation and GDMT can resolve massive biventricular thrombi, even in the presence of cerebral hemorrhage. Key lessons include: (1) Serial imaging guides dose titration; (2) Multidisciplinary input balances thromboembolic and bleeding risks; (3) High-altitude residency necessitates hemoglobin monitoring. Future studies should compare rivaroxaban vs. warfarin in similar cohorts.

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

Ethical approval was not required for the study involving humans in accordance with the local legislation and institutional requirements. Written informed consent to participate in this study was not required from the participants or the participants' legal guardians/next of kin in accordance with the national legislation and the institutional requirements. 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

CL: Writing – original draft. HL: Writing – original draft. ZC: Writing – original draft. TL: Resources, Writing – original draft. YW: Resources, Writing – original draft. YX: Writing – review & editing. RM: Writing – review & editing.

Funding

The author(s) declare that no financial support was received for the research and/or publication of this article.

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.

Generative AI statement

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Publisher's note

References

1. Hudec S, Hutyra M, Precek J, Latal J, Nykl R, Spacek M, et al. Acute myocardial infarction, intraventricular thrombus and risk of systemic embolism. *Biomed Pap Med Fac Univ Palacky Olomouc Czech Repub*. (2020) 164(1):34–42. doi: 10.5507/bp.2020.001

2. Levine GN, McEvoy JW, Fang JC, Ibeh C, McCarthy CP, Misra A, et al. Management of patients at risk for and with left ventricular thrombus: a scientific statement from the American Heart Association. *Circulation*. (2022) 146(15): e205–23. doi: 10.1161/CIR.0000000000001092

3. Kaya A, Hayıroğlu Mİ, Keskin M, Öz A, Aydın BA. Giant biventricular thrombi presenting with acute myocardial infarction. *Indian Heart J*. (2016) 68(Suppl 2): S153–5. doi: 10.1016/j.ihj.2016.03.006

4. Ozer M, Abbasi F, Mahdi M, Goksu SY, Struble E. Massive biventricular thrombi complicating new-onset heart failure in a patient with suspected COVID-19. *J Cardiol Cases*. (2021) 24(3):144–7. doi: 10.1016/j.jccase.2021.02.016

5. Paç FA, Cağdaş DN. Treatment of massive cardiac thrombi in a patient with protein C and protein S deficiency. *Blood Coagul Fibrinolysis*. (2007) 18(7):699–702. doi: 10.1097/MBC.0b013e3282eff788

6. Curiale A, Vallabhaneni S, Longo S, Shirani J. Massive left ventricular thrombosis in pericardial decompression syndrome. *Echocardiography*. (2021) 38(8):1471–3. doi: 10.1111/echo.15151

7. Dregoesc MI, Istrate M, Sandu I, Iancu AC. Massive biventricular thrombosis after transfemoral transcatheter aortic valve implantation. *Eur Heart J*. (2022) 43(16):1599. doi: 10.1093/eurheartj/ehac023

8. Barforoshi S, Sharim J, Budoff MJ. Massive biventricular thrombi in a patient with endometrial adenocarcinoma diagnosed on computed tomography angiography. *Clin Med Insights Cardiol*. (2023) 17:11795468231182762. doi: 10.1177/11795468231182762

9. Campisi S, Fuzellier JF, VolaGiant M, Favre JP. Left ventricular thrombus formation associated with heparin-induced thrombocytopenia. *Ann Thorac Surg*. (2014) 98(6):e143–5. doi: 10.1016/j.athoracsur.2014.08.054

10. Şeker T, Baykan AO, Börekçi A, Gür M, Çaylı M. Successful treatment of a huge thrombus with thrombolytic therapy in a patient with normal left ventricle function and Takayasu arteritis. *Turk Kardiyol Dern Ars*. (2014) 42(8):763–6. doi: 10.5543/tkda.2014.60687

11. Papadopoulos G, Nunez A, Grodman R, Mamadova A, Rahmany Z, El-Eshmawi A, et al. Massive mobile left ventricular thrombus in a patient with normal left ventricular systolic function. *CASE (Phila)*. (2019) 3:277–9. doi: 10.1016/j.case.2019.07.007

12. Hooks M, Okasha O, Velangi PS, Nijjar PS, Farzaneh-Far A, Shenoy C. Left ventricular thrombus on cardiovascular magnetic esonance imaging in nonischaemic cardiomyopathy. *Eur Heart J Cardiovasc Imaging*. (2021) 22(12):1425–33. doi: 10.1093/ehjci/jeaa244

13. Oh JK, Park JH, Lee JH, Kim J, Seong IW. Shape and mobility of a left ventricular thrombus are predictors of thrombus resolution. *Korean Circ J*. (2019) 49:829–37. doi: 10.4070/kcj.2018.0346

frontiers | Frontiers in Cardiovascular Medicine

TYPE Case Report
PUBLISHED 12 May 2025
DOI 10.3389/fcvm.2025.1538389

OPEN ACCESS

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RECEIVED 02 December 2024
ACCEPTED 22 April 2025
PUBLISHED 12 May 2025

CITATION
Wang Z, Shi Y, Chen Y and Chen L (2025) Case Report: Deep sternal wound infection caused by *Mycoplasma hominis* after cardiac surgery. Front. Cardiovasc. Med. 12:1538389. doi: 10.3389/fcvm.2025.1538389

Case Report: Deep sternal wound infection caused by *Mycoplasma hominis* after cardiac surgery

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Deep sternal wound infection (DSWI) caused by *Mycoplasma hominis* after cardiac surgery is very rare and easily overlooked. This paper reports the case of a patient with DSWI after cardiac surgery. The patient had an unexplained fever postoperatively. On the seventh day after surgery, the incision opened, and there was pus leakage. On the 11th day after surgery, the pus culture indicated *Mycoplasma hominis* infection. The patient was cured after treatment with omadacycline and secondary surgery.

KEYWORDS

***Mycoplasma hominis*, deep mediastinitis, cardiac surgery, omadacycline, DSWI**

Introduction

Deep sternal wound infection (DSWI) is one of the most serious complications after cardiac surgery through the sternum, with an incidence rate worldwide ranging from 0.5% to 5.6% (1). Patient risk factors include female gender, age, diabetes, renal failure, smoking, obesity, breast size, steroid use, and chronic obstructive pulmonary disease (2, 3). Surgery-related risk factors include surgery duration, internal mammary artery use, re-thoracotomy, blood product use, intensive care unit stay, and duration of mechanical ventilation (4, 5). The most common pathogens are Methicillin-sensitive *Staphylococcus aureus* (45%), Methicillin-resistant *S. aureus* (16%), Gram-negative bacilli (17%), Coagulase-negative Staphylococci (13%), and Streptococci (5%) (6). *Mycoplasma hominis* is a rare pathogen for DSWI, with less than 20 cases of postcardiac surgery *M. hominis* mediastinitis reported globally in the past 50 years (6, 7). This paper reports a case of a patient who underwent valvular replacement and coronary artery bypass grafting and presented with fever and chest pain postoperatively. *M. hominis* was found in the incision pus culture. The patient showed improvement after standardized anti-infection treatment and pectoralis major muscle flap transfer and was discharged in good condition.

Case presentation

A 59-year-old man was admitted to the hospital for "chest tightness and shortness of breath after exertion for 1 month." He had a history of "cerebral infarction" and underwent coronary stent implantation 1 year ago at a hospital in another underdeveloped province about 2,000 km far away. He came to our hospital for medical treatment through a health assistance project between the two provincial governments. He denied any other medical history. Echocardiography in our hospital revealed moderate stenosis with moderate insufficiency of the aortic valve. The ejection fraction (EF)% value was 68%.

Coronary angiography indicated 85% stenosis in the middle of the right coronary artery and 80%–90% stenosis in the distal segment of the left circumflex branch. The patient's predicted mortality was 1.29% based on EuroSCORE II. The calculated additive EuroSCORE II value was 4.333.

The patient underwent mechanical aortic valve replacement combined with coronary artery bypass grafting on 26 September 2024. Two graft procedures were performed: a saphenous vein sequential graft, harvested from the right thigh, was anastomosed from the ascending aorta to the left circumflex coronary artery and subsequently to the posterior descending branch of the right coronary artery.

The total surgical time was 380 min, of which the cardiopulmonary bypass time was 127 min. The aortic cross-clamp time during surgery was 180 min.

After the patient was admitted to the surgical intensive care unit (SICU) after surgery, the SICU nurse reported that he had a swelling of the foreskin and several small ruptures in the local area (unfortunately, the photographs taken at that time are missing now). Local disinfection and foreskin reduction were immediately performed, and the swelling gradually reduced.

The patient had a fever on postoperation day (POD) 1, with the highest temperature reaching 39.4°C on POD 5. It was not until POD 11 that the body temperature returned to normal (Figure 1). The patient's white blood cell count and the percentage of neutrophils were slightly elevated. Procalcitonin levels decreased over time, which was considered to have no guiding significance for *M. hominis* infection. C-reactive protein (CRP) levels changed with time and treatment measures (Figure 2).

The urinary catheter was removed on POD 7, and the central venous catheter was removed on POD 8. Chest CT scans performed on POD 1 revealed mild consolidation in the bilateral lower lung lobes and minimal pleural effusion, with subsequent imaging on POD 6 demonstrating improvement in these findings; no mediastinal abnormalities were detected in either scan.

On POD 7 (3 October 2024), the patient's incision was dehisced, revealing a large amount of yellow-white pus exuding, with poor healing of subcutaneous tissue and sternum (Figure 3).

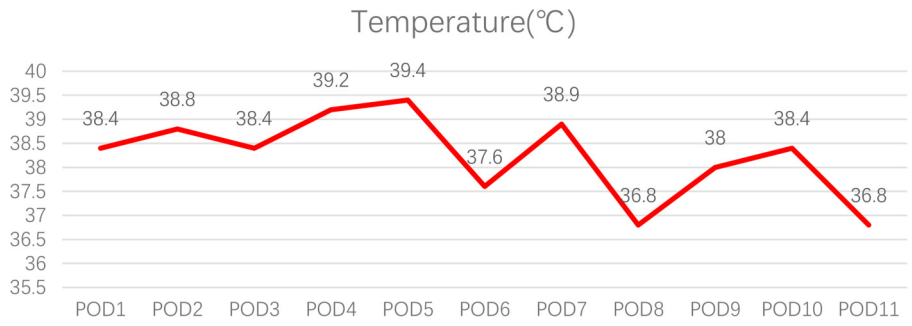

FIGURE 1
Postoperative changes in temperature.

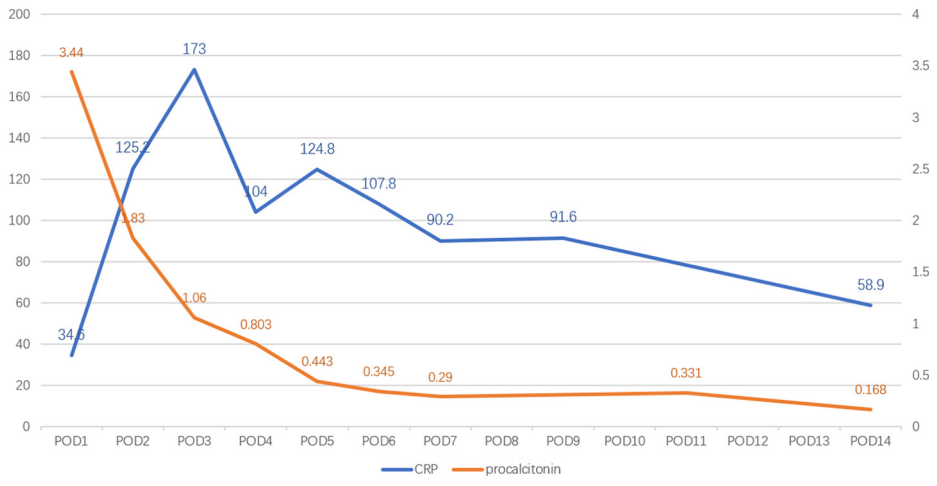

FIGURE 2
Postoperative changes in CRP (mg/L) and procalcitonin (ng/ml).

FIGURE 3
Incision dehiscence.

FIGURE 4
Cultivation results.

Before sternal dehiscence, localized erythema and tenderness around the incision were observed on POD 3. However, these signs were initially attributed to routine postoperative inflammation without special attention.

Multiple continuous samplings were taken from the incision for culture since sternal dehiscence. Finally, on POD 11, the first positive culture result came out, which indicated *M. hominis* (Figure 4). An antibiogram was not performed due to technical limitations in *M. hominis* susceptibility testing, which is rarely conducted in clinical practice, and the absence of institutional infrastructure for such specialized assays. After a multidisciplinary team discussion, Omadacycline was administered for anti-infection treatment. After using the medication, the incision condition gradually improved, and no positive results were cultured again. On POD 21, an open chest debridement and bilateral pectoralis major muscle flap plugging surgery was successfully performed. The patient was discharged 14 days after the secondary surgery.

The anti-infection treatment is detailed in Table 1.

At 1-, 3-, and 5-month follow-ups, the patient exhibited no fever recurrence, with complete sternal wound healing confirmed on physical examination. Cardiac function remained normal, as evidenced by transthoracic echocardiography and electrocardiography. In chest CT scans, no mediastinal abnormalities were observed. Laboratory assessments revealed normalized inflammatory markers and stable coagulation profiles (international normalized ratio: 1.8–2.5 under warfarin therapy). The patient resumed full-time work and daily activities without functional limitations.

The patient initially expressed frustration due to incision dehiscence and delayed diagnosis. Through detailed communication and empathic counseling, his understanding and adherence improved. At discharge, he reported high satisfaction with the care team's efforts and gratitude for the successful outcome.

Discussion

DSWI is a rare but potentially devastating complication of median sternotomy performed in cardiac surgery. Risk factors can be broadly divided into preoperative, intraoperative, and postoperative factors, including female sex, obesity, diabetes mellitus, smoking and chronic obstructive pulmonary disease (COPD), bilateral internal mammary artery harvesting, prolonged cardiopulmonary bypass time, and re-exploration for bleeding. In this case, the patient exhibited the following DSWI risks: smoking and single internal mammary artery harvesting. The risk factors had limited guiding significance for this patient.

M. hominis infection causing DSWI after cardiac surgery is very rare and difficult to diagnose. The main reason is that *M. hominis* has high requirements for culture, and ordinary culture methods make it difficult to obtain positive results (8). Currently, many studies have adopted molecular diagnosis and genomic sequencing methods to diagnose *Mycoplasma* infections (6, 7) and have achieved good results, but new technology means high costs and low popularity. So, conventional culture and testing remain the main methods for grassroots hospitals to find the causative bacteria of infection. First, qualified test specimens and multiple tests may increase the positive detection rate. Second,

TABLE 1 Anti-infection treatment detail.

Mediation	Dosage (g)	Frequency of use	Start time	End time
Cefuroxime	1.5	Every 8 h	26 September 2024 16:18	28 September 2024 09:33
Piperacillin–tazobactam	4.5	Every 8 h	28 September 2024 09:33	30 September 2024 08:50
Meropenem	1	Every 8 h	30 September 2024 08:50	2 October 2024 10:50
Piperacillin–tazobactam	4.5	Every 8 h	3 October 2024 09:22	17 October 2024 09:07
Omadacycline	0.1	Everyday	7 October 2024 16:38	29 October 2024 09:22
Meropenem	1	Every 8 h	17 October 2024 18:03	25 October 2024 09:10
Vancomycin	1	Every 12 h	17 October 2024 18:03	25 October 2024 09:10

culture dishes with no bacterial growth within 48 h should not be directly reported as negative and discarded. Previous studies have proven that *M. hominis* requires at least 4 days or even longer under conventional culture to appear in colonies (9).

Many articles also mention an interesting phenomenon about *M. hominis*, which is that all patients with DSWI caused by postoperative *M. hominis* are male. *M. hominis* mainly colonizes in the human respiratory and urinary tract and, because of the anatomical differences between male and female urethras, the damage to the male urethra from catheterization is much greater than that to the female urethra. Therefore, many researchers believe that *Mycoplasma* infection is related to urethral injury. In this case, the patient did indeed have penile edema and local skin ulceration on the first and second days after surgery. In addition, the patient's graft vessel for the bypass was taken from the great saphenous vein of the right thigh, which is close to the perineal area. It is possible that strict aseptic operation was not maintained, leading to *Mycoplasma* infection. Therefore, perioperative perineal cleaning, preoperative catheterization operations, and strict aseptic operations during surgery are crucial for preventing infection.

In this case, the patient was treated with piperacillin–sulbactam and meropenem without therapeutic effect before the pathogen was identified. After the culture confirmed a *Mycoplasma* infection, omadacycline was used for anti-infection treatment. The decision to initiate Omadacycline was guided by a multidisciplinary team consultation involving infectious disease and emergency medicine specialists. Key factors included the following: confirmed *M. hominis* infection with suspected genitourinary origin, given the patient's postoperative penile edema and proximity of the saphenous vein graft harvest site to the perineal region; *in vitro* evidence demonstrating the potent activity of Omadacycline against *Mycoplasma genitalium* [minimal inhibit concentration (MIC) $\leq$0.5 mg/ml], even in isolates resistant to other tetracyclines (10); and pharmacokinetic advantages, including intravenous administration for rapid therapeutic effect, and broad-spectrum coverage against Gram-positive and Gram-negative pathogens, which mitigated the risks of polymicrobial infection.

Omadacycline is a tetracycline-class antibiotic that inhibits bacterial protein synthesis by binding to the 30S ribosomal subunit. It has antibacterial activity against aerobic Gram-positive bacteria such as *Enterococcus faecalis*, *Enterococcus faecium*, vancomycin-resistant enterococci, methicillin-resistant *S. aureus*, various streptococci, *Corynebacterium jeikeium*, and Enterobacteriaceae resistant to ceftazidime and carbapenems, as well as atypical pathogens (11). In previous case reports on anti-infection treatment with quinolones combined with doxycycline, with or without decortication surgery, 28.6% of patients died due to uncontrollable infection (6).

This case highlights the critical role of persistent culturing with prolonged incubation to isolate *M. hominis*, a fastidious pathogen rarely detected by conventional methods. To our knowledge, it is the first reported use of omadacycline for successfully treating DSWI caused by *M. hominis* after cardiac surgery, addressing a literature gap. Limitations include the single-center, single-case design and the inability to perform susceptibility testing due to institutional constraints, underscoring challenges in managing atypical infections in resource-limited settings.

Prospective multicenter studies with larger cohorts are warranted to validate the efficacy of omadacycline in DSWI caused by *M. hominis*, while advancing rapid molecular diagnostics and standardized susceptibility protocols could enhance the management of such rare, challenging infections.

Summary

M. hominis-induced deep sternal wound infection is a relatively rare complication after cardiac surgery, which is considered to be related to catheterization. *M. hominis* is difficult to culture and, therefore, molecular testing or genomic sequencing methods can be used to assist in diagnosis for units with the capability. For confirmed *M. hominis* infections in DSWI, omadacycline can be used for anti-infection treatment.

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 humans were approved by the ethics committee of Dongyang People's Hospital. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation in this study was provided by the participants' legal guardians/next of kin. 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

ZW: Conceptualization, Data curation, Formal analysis, Funding acquisition, Writing – original draft, Writing – review & editing. YS: Software, Supervision, Validation, Visualization, Writing – original draft. YC: Investigation, Methodology, Project administration, Resources, Writing – original draft. LC: Conceptualization, Data curation, Investigation, Methodology, Software, Supervision, Writing – original draft, Writing – review & editing.

Funding

The author(s) declare that no financial support was received for the research and/or publication of this article.

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. Vos RJ, Van Putte BP, Kloppenburg GTL. Prevention of deep sternal wound infection in cardiac surgery: a literature review. *J Hosp Infect.* (2018) 100:411–20. doi: 10.1016/j.jhin.2018.05.026

2. Lepelletier D, Bourigault C, Roussel JC, Lasserre C, Leclère B, Corvec S, et al. Epidemiology and prevention of surgical site infections after cardiac surgery. *Med Mal Infect.* (2013) 43:403–9. doi: 10.1016/j.medmal.2013.07.003

3. Leung Wai Sang S, Chaturvedi R, Alam A, Samoukovic G, de Varennes B, Lachapelle K. Preoperative hospital length of stay as a modifiable risk factor for mediastinitis after cardiac surgery. *J Cardiothorac Surg.* (2013) 8:45. doi: 10.1186/1749-8090-8-45

4. Diez C, Koch D, Kuss O, Silber R-E, Friedrich I, Boergermann J. Risk factors for mediastinitis after cardiac surgery—a retrospective analysis of 1700 patients. *J Cardiothorac Surg.* (2007) 2:23. doi: 10.1186/1749-8090-2-23

5. Lu JCY, Grayson AD, Jha P, Srinivasan AK, Fabri BM. Risk factors for sternal wound infection and mid-term survival following coronary artery bypass surgery. *Eur J Cardiothorac Surg.* (2003) 23:943–9. doi: 10.1016/s1010-7940(03)00137-4

6. Trouillet J-L, Vuagnat A, Combes A, Bors V, Chastre J, Gandjbakhch I, et al. Acute poststernotomy mediastinitis managed with debridement and closed-drainage aspiration: factors associated with death in the intensive care unit. *J Thorac Cardiovasc Surg.* (2005) 129:518–24. doi: 10.1016/j.jtcvs.2004.07.027

7. Le Guern R, Loïez C, Loobuyck V, Rousse N, Courcol R, Wallet F. A new case of Mycoplasma hominis mediastinitis and sternal osteitis after cardiac surgery. *Int J Infect Dis.* (2015) 31:53–5. doi: 10.1016/j.ijid.2014.12.028

8. Houpikian P, Raoult D. Blood culture-negative endocarditis in a reference center: etiologic diagnosis of 348 cases. *Medicine.* (2005) 84:162–73. doi: 10.1097/01.md.0000165658.82869.17

9. Filthuth I, Emler S, Jacobs E, Auckenthaler R. Isolation of *Mycoplasma hominis* on CDC anaerobic blood agar. *Eur J Clin Microbiol Infect Dis.* (1996) 15:896–7. doi: 10.1007/BF01691229

10. Waites KB, Crabb DM, Atkinson TP, Geisler WM, Xiao L. Omadacycline is highly active *in vitro* against *Mycoplasma genitalium*. *Microbiol Spectr.* (2022) 10: e0365422. doi: 10.1128/spectrum.03654-22

11. Hussar DA, Chahine EB. Omadacycline tosylate, sarecycline hydrochloride, rifamycin sodium, and moxidectin. *J Am Pharm Assoc.* (2019) 59:756–60. doi: 10.1016/j.japh.2019.07.016

frontiers | Frontiers in Cardiovascular Medicine

TYPE Case Report
PUBLISHED 27 May 2025
DOI 10.3389/fcvm.2025.1511221

OPEN ACCESS

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RECEIVED 14 October 2024
ACCEPTED 07 May 2025
PUBLISHED 27 May 2025

CITATION
Tian Y, Huang E, Zhang M, Fan J, Li W, Yang X, Su W and Deng X (2025) Case Report: Minimally invasive management of suspected active bleeding from intercostal vessel after axillary thoracotomy ventricular septal defect repair: an application of Foley catheter. Front. Cardiovasc. Med. 12:1511221. doi: 10.3389/fcvm.2025.1511221

Case Report: Minimally invasive management of suspected active bleeding from intercostal vessel after axillary thoracotomy ventricular septal defect repair: an application of Foley catheter

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The right axillary thoracotomy, an alternative approach for selected open-heart procedures, offers aesthetic advantages. However, intercostal vessel injury is a potential postoperative complication that can lead to major bleeding. Herein, we report a case of postoperative active bleeding from intercostal vessel injury after right axillary thoracotomy for ventricular septal defect repair. Hemorrhage was successfully halted by compressing the suspected bleeding site with a Foley catheter inserted through the chest wall. This case demonstrates a simple management method of active intercostal vascular bleeding after cardiac operation. It may be an appropriate option in selected postoperative patients with a high index of suspicion of intercostal vascular bleeding after operation via a thoracotomy approach.

KEYWORDS

Foley catheter, chest drain, postoperative bleeding, ventricular septal defect, minimally invasive, axillary thoracotomy

Introduction

Recent years have witnessed the advent of minimally invasive surgical techniques, including interventional procedures, robotics, and axillary approaches. The right axillary thoracotomy is an alternative approach for selected cardiac procedures requiring cardiopulmonary bypass. This technique offers an aesthetic advantage since the incision is hidden beneath the arm (1, 2). However, intercostal vessel injury is a potential postoperative complication that can lead to major bleeding. The conventional management of active hemorrhage involves open exploration to locate and repair the bleeding site. This approach, while effective, is invasive and can be associated with higher morbidity and longer recovery times.

Achieving hemostasis through balloon compression has rarely been documented. We present a case of active intercostal bleeding following a right axillary thoracotomy, which was controlled by inserting a Foley catheter to compress the presumed bleeding site. This minimally invasive technique offers a less invasive alternative to conventional

re-exploration, potentially reducing morbidity and recovery time. We have reported this case in line with the SCARE criteria (3–5).

Presentation of case

A 5-year-old child was admitted to our institution for repair of a ventricular septal defect (VSD). The operation was performed under general anesthesia and extracorporeal circulation via a right axillary thoracotomy at the fourth intercostal space. Following closure of the VSD, intermittent 2:1 conduction was observed upon removing the cross clamp. A temporary pacing wire was inserted into the right ventricle and passed through the sixth intercostal space. During chest closure, active bleeding was noted at the site where the pacing wire penetrated the chest wall. Prompt action was taken, including the placement of a purse-string suture to achieve hemostasis. Subsequent verification of the surgical and drain incision sites confirmed no further active bleeding.

Upon returning to the ICU following surgery, the patient exhibited a pale complexion and lips. The heart rate was alarmingly high, ranging from 140 to 160 beats per minute, while blood pressure showed significant instability, fluctuating between 53 and 60 mmHg systolic and 32–36 mmHg diastolic. Notably, there was substantial drainage from the right pleural drainage tube, with approximately 260 ml collected within the first three hours post-operation. Concurrently, the central venous pressure (CVP) measured 4 cmH2O, and the urine output was approximately 35 ml per hour.

Based on the clinical manifestations of low blood pressure, elevated heart rate, and substantial thoracic drainage volume, along with the observation that arterial blood typically exhibits a jet-like spurt pattern while the drainage fluid appears bright red with relatively slow bleeding velocity and low pressure, we hypothesize that the persistent hemorrhage may result from intercostal venous vascular injury caused by pacemaker lead penetration, leading to continuous bleeding.This was despite a meticulous hemostatic examination during the closure of the chest cavity. Considering the patient's age and the technical challenges associated with quickly locating and suturing the bleeding site, re-exploring the chest could potentially increase the risk of secondary injury. In light of these considerations, we promptly prepared for bedside exploration and opted for a Foley catheter to achieve compression hemostasis in the meantime. Selecting a 10-French Foley catheter based on the patient's age, we infused 5 ml of normal saline as per the catheter's specifications. The catheter was then gently withdrawn and clamped against the chest wall to effectively compress the bleeding site (see Figure 1). This intervention led to a gradual reduction in the drainage volume. Subsequent echocardiography and bedside chest radiography were conducted to rule out the possibility of cardiac tamponade resulting from obstruction of the drainage tube. The patient's heart rate stabilized between 100 and 140 beats per minute, and blood pressure improved to a stable range of 80–90 mmHg systolic and 50–60 mmHg diastolic. The 24-h drainage volume decreased to approximately 20 ml, with CVP values between 5 and 8 cmH2O, red blood cell count (RBC) at $4.36 \times 106/m^3$, hemoglobin (HGB) at 128 g/L, and platelet count (PLT) at $150{,}000/m^3$. These vital signs indicated that the Foley catheter had successfully compressed the bleeding site.

After confirming the absence of active bleeding and the restoration of sinus rhythm, the drainage tube and pacemaker lead were safely removed on the second postoperative day. At the three-month follow-up, repeat imaging revealed no significant abnormalities, and the child's overall condition was favorable. Informed consent for the publication of this case was obtained from the parents.

Discussion

Open heart surgery via right axillary thoracotomy is becoming the choice of approach in selected cases (6, 7). The utilization of a right axillary thoracotomy for certain cardiac procedures offers advantages such as reduced trauma and quicker recovery times compared to median sternotomy (8). However, it is also associated with specific risks, including an increased likelihood of intercostal vessel injury and subsequent bleeding due to the proximity of the incision to these structures.

The placement of chest drains tubes following open heart surgery is standard practice. Careful consideration was given to the placement of the drain, with efforts made to puncture along the upper edge of the lower rib, followed by meticulous checks to ensure no active bleeding at the puncture site.

The insertion of temporary epicardial pacing wires can further complicate matters, especially in cases where positioning the pacing lead proves challenging. While pacing leads are not universally utilized in all cardiac procedures, they are employed in scenarios similar to the one presented in this case report. Not like the placement of a chest drain, it is difficult to confirm, in relation to the intercostal vessels, the site where the lead penetrates inside out through the chest wall, hence increasing the risk of injury and bleeding of intercostal vessels, which was the case for the present patient.

Swift control of hemorrhage is critical following vascular injury to restore blood volume and prevent hemodynamic compromise. External compression of the wound represents a rapid means of achieving hemostasis and limiting ongoing blood loss. The insertion of a Foley catheter with subsequent inflation of the balloon can effectively tamponade bleeding from an injured intercostal vessel through direct compression at the hemorrhage site. The selection of the 10-French catheter was based on three main factors: 1. Pediatric intercostal space dimensions, which average 4–6 mm in 5-year-olds. 2. Balloon inflation capacity, with 5 ml providing optimal radial pressure. 3. Minimizing trauma while ensuring effective tamponade. For adult applications, we would recommend upsizing to 12–14-French catheters with proportionally larger balloon volumes (8–10 ml).

In this case, while re-exploration was contemplated, it was not immediately pursued to minimize trauma to the patient. Instead, a presumption of the hemorrhage site was made based on clinical evidence. The surgical incision and drain port had been confirmed with no bleeding, yet the pacing lead site was found with bleeding during chest closure even though hemostasis was achieved at once. Therefore, the postoperative bleeding was highly suspected to be

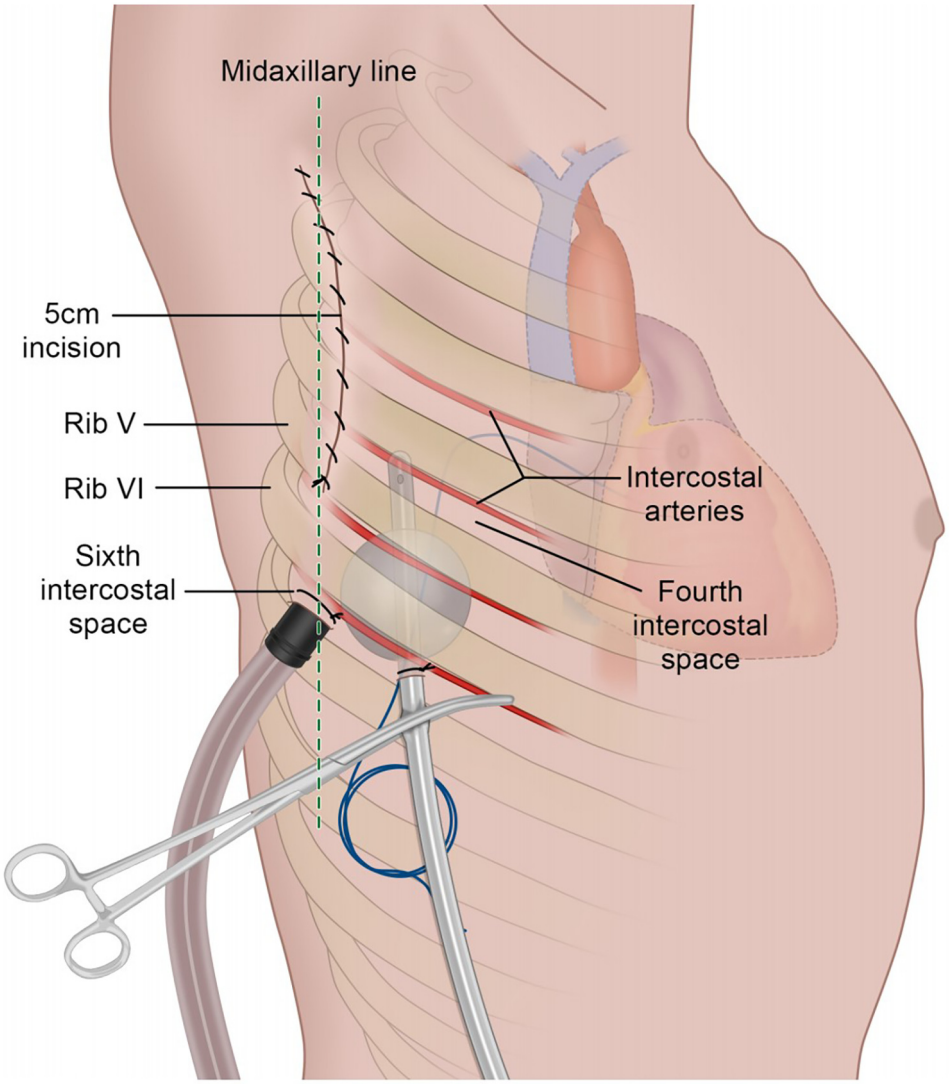

FIGURE 1
Foley catheter compresses the bleeding site.

from the pacing lead site. Balloon tamponade was successfully employed to achieve hemostasis without the need for further surgical intervention. This minimally invasive approach not only effectively controlled bleeding but also minimized additional trauma and expedited the patient's recovery. We have compiled a comparative table summarizing this minimally invasive approach, which is presented as Table 1 at the end of the article.

Similar applications of balloon compression for damage control have been documented across various medical specialties, including in cases of hepatic, abdominal, orthopedic, and previous cardiac surgical trauma, and multiple heel and rib fractures and multiple rib fractures, and similar to the present case for bleeding from intercostal vascular injuries after cardiac surgery (9–14).

This case highlights the critical role of vigilant postoperative monitoring and the strategic application of innovative techniques, such as Foley catheter compression, in effectively managing complications like active intercostal bleeding following cardiac surgery via a thoracotomy approach. To further enhance the clarity and applicability of our methodology, we have added a new flowchart detailing the patient selection criteria as follows Figure 2. Figure 2 flowchart detailing the patient selection for balloon compression using a Foley's catheter.

Conclusion

Our report highlights a straightforward management approach for active intercostal vascular bleeding following cardiac surgery. This method may represent a suitable option in carefully selected postoperative patients where there is a high suspicion of intercostal vascular bleeding via a thoracotomy approach. By employing balloon tamponade, clinicians can effectively address hemorrhage while minimizing the need for reoperation, thereby promoting patient recovery and reducing the risk of further complications.

TABLE 1 Comparison of different methods for intercostal vessel injury management.

Management option	Indication	Procedure description	Advantages	Risks/complications
Foley Catheter (intercostal compression)	Small to moderate bleeding in stable patients	Insert Foley catheter into pleural space to provide localized compression.	Non-invasive, bedside, minimal anesthesia.	Displacement, infection, incomplete hemostasis.
Surgical Re-exploration	Severe or uncontrolled bleeding	Thoracotomy for direct visualization and vessel ligation/cauterization.	Definitive control; manages complex injuries.	Infection, bleeding, lung damage, prolonged recovery.
Endovascular techniques	Moderate to severe bleeding, high-risk surgical patients	Embolization or stenting to occlude bleeding vessels.	Minimally invasive, effective for unstable patients.	Embolization failure, re-bleeding, catheter-related issues.
Chest tube with hemostasis	Moderate bleeding with hemothorax	Chest tube inserted for blood evacuation and clot formation.	Simple, helps control bleeding while monitoring.	Recurrent bleeding, infection, tube dislodgement.

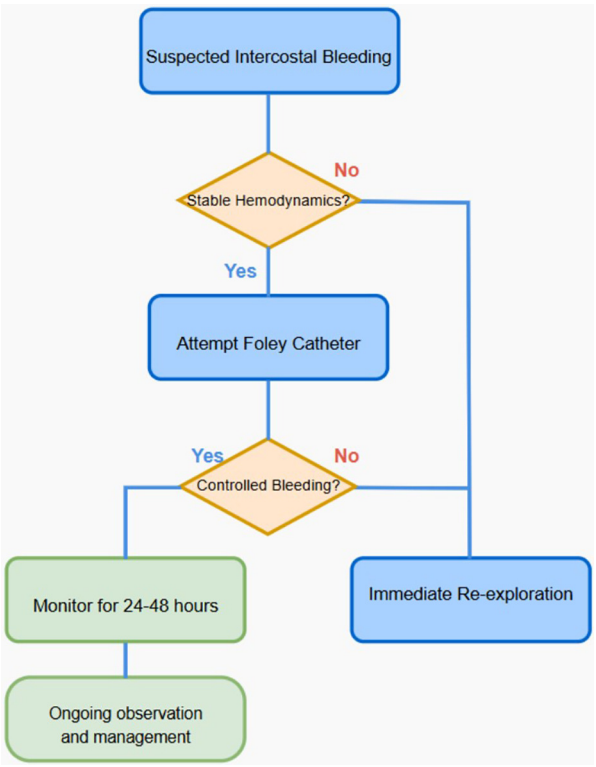

FIGURE 2
Evaluating the procedure of balloon compression using a Foley catheter.

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 authors.

Ethics statement

The studies involving humans were approved by Ethics Committee of Hunan Provincial Children's Hospital. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation in this study was provided by the participants' legal guardians/next of kin. Written informed consent was obtained from the individual(s), and minor(s)' legal guardian/next of kin, for the publication of any potentially identifiable images or data included in this article.

Author contributions

YT: Writing – original draft, Writing – review & editing, Methodology. EH: Writing – original draft, Writing – review & editing. MZ: Writing – original draft, Writing – review & editing, Methodology. JF: Writing – original draft, Writing – review & editing, Methodology. WL: Writing – original draft, Writing – review & editing, Methodology. XY: Writing – original draft, Writing – review & editing, Methodology. WS: Writing – original draft, Writing – review & editing, Methodology XD: Writing – original draft, Writing – review & editing, Funding acquisition, Investigation, Supervision.

Funding

The author(s) declare that no financial support was received for the research and/or publication of this article.

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. Alicandri-Ciufelli M, Maccarrone F, Botti C, Pavesi G, Presutti L. Use of Foley catheter in control of internal carotid hemorrhage during endoscopic endonasal surgery. *Folia Med (Plovdiv)*. (2021) 63(5):809–14. doi: 10.3897/folmed.63.e56461

2. An K, Li S, Yan J, Wang X, Hua Z. Minimal right vertical infra-axillary incision for repair of congenital heart defects. *Ann Thorac Surg*. (2022) 113(3):896–902. doi: 10.1016/j.athoracsur.2021.01.052

3. Apaydin AZ, Posacioglu H, Islamoglu F, Telli A. A practical tool to control bleeding during sternal reentry for pseudoaneurysm of the ascending aorta. *Ann Thorac Surg*. (2003) 75(3):1037–8. doi: 10.1016/S0003-4975(02)04369-2

4. Atalay A, Yilmaz M, Turkcan BS, Ecevit AN, Ozler B, Azak E, et al. Can right infra-axillary vertical thoracotomy make a big difference in surgical technique preference? *Heart Lung Circ*. (2022) 31(10):1419–24. doi: 10.1016/j.hlc.2022.06.661

5. Beysard N, Pasquier M, Zingg T, Carron PN, Darioli V. The use of foley catheter tamponade for bleeding control in penetrating injuries. *ScandJ Trauma Resusc Emerg Med*. (2021) 29(1):165. doi: 10.1186/s13049-021-00975-2

6. Dodge-Khatami J, Dodge-Khatami A. Advantages of a mini right axillary thoracotomy for congenital heart defect repair in children. *Cardiol Young*. (2022) 32(2):276–81. doi: 10.1017/S1047951121001979

7. Dodge-Khatami J, Noor R, Riggs KW, Dodge-Khatami A. Mini right axillary thoracotomy for congenital heart defect repair can become a safe surgical routine. *Cardiol Young*. (2023) 33(1):38–41. doi: 10.1017/S1047951122000117

8. Elmoghrabi A, Mohamed M, Eggers A, Parmar N, McCann M. More than just a urinary catheter—haemorrhage control by using a foley catheter in a penetrating aortic root injury. *Trauma Case Rep*. (2016) 5:13–7. doi: 10.1016/j.tcr.2016.09.006

9. Galea J, Manche A. Method of hemorrhage control from the aorta after repair of a dissected aortic aneurysm. *Ann Thorac Surg*. (2017) 103(3):e299–300. doi: 10.1016/j.athoracsur.2016.08.105

10. Hong ZN, Chen Q, Lin ZW, Zhang GC, Chen LW, Zhang QL, et al. Surgical repair via submammary thoracotomy, right axillary thoracotomy and median sternotomy for ventricular septal defects. *J Cardiothorac Surg*. (2018) 13(1):47. doi: 10.1186/s13019-018-0734-5

11. Hsu TC. Foley balloons ' tamponade is an efective method for controlling massive pelvic bleeding during colorectal surgery. *Ann Coloproctol*. (2022) 38(1):69–71. doi: 10.3393/ac.2020.00682.0097

12. Kong V, Ko J, Cheung C, Lee B, Leow P, Thirayan V, et al. Foley catheter balloon tamponade for actively bleeding wounds following penetrating neck injury is an efective technique for controlling non-compressible junctional external haemorrhage. *World J Surg*. (2022) 46(5):1067–75. doi: 10.1007/s00268-022-06474-4

13. Lu YM, Guo YR, Zhou MY, Wang Y. Indwelling intrauterine Foley balloon catheter for intraoperative and postoperative bleeding in cesarean scar pregnancy. *J Minim Invasive Gynecol*. (2020) 27(1):94–9. doi: 10.1016/j.jmig.2019.02.024

14. Ruiz-Tovar J, Priego-Jimenez P, Paiva-Coronel GA. Use of foley 's catheter to control port-site bleeding in bariatric surgery. *Obes Surg*. (2012) 22(2):306–8. doi: 10.1007/s11695-011-0572-1

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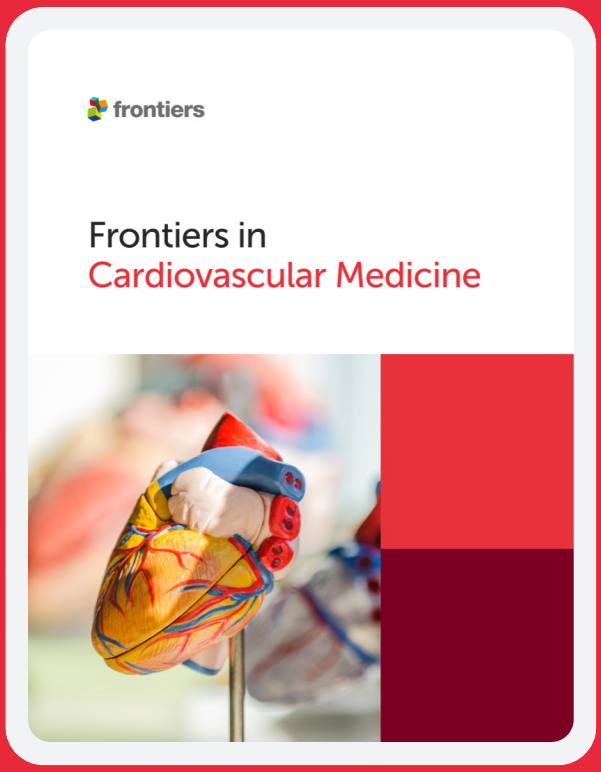

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