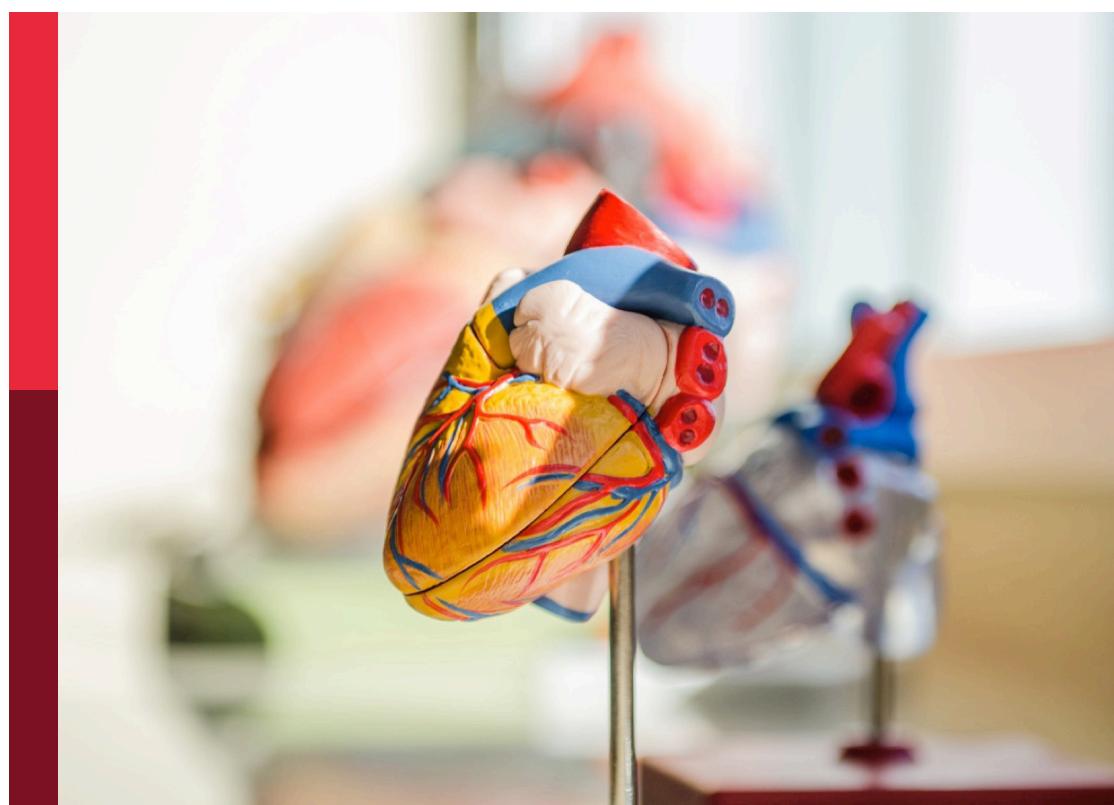


Case reports in heart surgery

2023

Edited by
Giuseppe Gatti

Published in
Frontiers in Cardiovascular Medicine



FRONTIERS EBOOK COPYRIGHT STATEMENT

The copyright in the text of individual articles in this ebook is the property of their respective authors or their respective institutions or funders. The copyright in graphics and images within each article may be subject to copyright of other parties. In both cases this is subject to a license granted to Frontiers.

The compilation of articles constituting this ebook is the property of Frontiers.

Each article within this ebook, and the ebook itself, are published under the most recent version of the Creative Commons CC-BY licence. The version current at the date of publication of this ebook is CC-BY 4.0. If the CC-BY licence is updated, the licence granted by Frontiers is automatically updated to the new version.

When exercising any right under the CC-BY licence, Frontiers must be attributed as the original publisher of the article or ebook, as applicable.

Authors have the responsibility of ensuring that any graphics or other materials which are the property of others may be included in the CC-BY licence, but this should be checked before relying on the CC-BY licence to reproduce those materials. Any copyright notices relating to those materials must be complied with.

Copyright and source acknowledgement notices may not be removed and must be displayed in any copy, derivative work or partial copy which includes the elements in question.

All copyright, and all rights therein, are protected by national and international copyright laws. The above represents a summary only. For further information please read Frontiers' Conditions for Website Use and Copyright Statement, and the applicable CC-BY licence.

ISSN 1664-8714
ISBN 978-2-8325-5809-6
DOI 10.3389/978-2-8325-5809-6

About Frontiers

Frontiers is more than just an open access publisher of scholarly articles: it is a pioneering approach to the world of academia, radically improving the way scholarly research is managed. The grand vision of Frontiers is a world where all people have an equal opportunity to seek, share and generate knowledge. Frontiers provides immediate and permanent online open access to all its publications, but this alone is not enough to realize our grand goals.

Frontiers journal series

The Frontiers journal series is a multi-tier and interdisciplinary set of open-access, online journals, promising a paradigm shift from the current review, selection and dissemination processes in academic publishing. All Frontiers journals are driven by researchers for researchers; therefore, they constitute a service to the scholarly community. At the same time, the *Frontiers journal series* operates on a revolutionary invention, the tiered publishing system, initially addressing specific communities of scholars, and gradually climbing up to broader public understanding, thus serving the interests of the lay society, too.

Dedication to quality

Each Frontiers article is a landmark of the highest quality, thanks to genuinely collaborative interactions between authors and review editors, who include some of the world's best academicians. Research must be certified by peers before entering a stream of knowledge that may eventually reach the public - and shape society; therefore, Frontiers only applies the most rigorous and unbiased reviews. Frontiers revolutionizes research publishing by freely delivering the most outstanding research, evaluated with no bias from both the academic and social point of view. By applying the most advanced information technologies, Frontiers is catapulting scholarly publishing into a new generation.

What are Frontiers Research Topics?

Frontiers Research Topics are very popular trademarks of the *Frontiers journals series*: they are collections of at least ten articles, all centered on a particular subject. With their unique mix of varied contributions from Original Research to Review Articles, Frontiers Research Topics unify the most influential researchers, the latest key findings and historical advances in a hot research area.

Find out more on how to host your own Frontiers Research Topic or contribute to one as an author by contacting the Frontiers editorial office: frontiersin.org/about/contact

Case reports in heart surgery: 2023

Topic editor

Giuseppe Gatti — Azienda Sanitaria Universitaria Giuliano Isontina, Italy

Citation

Gatti, G., ed. (2024). *Case reports in heart surgery: 2023*.
Lausanne: Frontiers Media SA. doi: 10.3389/978-2-8325-5809-6

Table of contents

05 **Editorial: Case reports in heart surgery: 2023**
Giuseppe Gatti

08 **Case report: pulmonary artery perforation during transseptal puncture for left atrial appendage closure requires emergency cardiac operation**
Yue Wang, Beibei Song, Bing Liu, Hui Zhang, Chenglong Bi, Wenhao Liu, Gang Ma and Bo Li

14 **Case Report: Myocardial dissection caused by ruptured sinus of Valsalva aneurysm in association with a bicuspid aortic valve**
Xinyan Zhou, Yan Xu, Qian He, Na Tan, Jixiang Chu, Bin Liu, Yu Zhu, Chengde Liao and Yu Jiang

20 **Case Report: Right atrial mass arising from the Eustachian valve**
Jalal Jolou, Jérôme Martineau, Hajo Müller, Mustafa Cikirkcioglu and Christoph Huber

24 **Case Report: Pericardial patch repair of mitral annulus and mitral valve for a left atrial dissection caused by parasitic infective endocarditis**
PeiShan Chu, Yi Tang, XinPei Liu and Qi Miao

28 **Case Report: Surgical management of idiopathic pulmonary aneurysms and review surgical approaches**
Kui Wu, Xuan Fan, Xuanyi Hu, Xuejun Li and Siyuan Yang

34 **Case Report: Acute cerebral infarction caused by left atrial and right ventricular myxoma received emergency operation**
Chengbin Tang, Xianglong Gao, Tao Chen, Jun Shao, Tao Zhu, Xucai Zheng and Chuanli Ren

38 **Case Report: Incidental finding of an atresia of the inferior vena cava—a challenge for cardiac surgery**
Joscha Buech, Caroline Radner, Thomas Fabry, Simon Rutkowski, Christian Hagl, Sven Peterss and Maximilian A. Pichlmaier

41 **Case Report: Giant left atrial cystic tumor: myxoma or intracardiac blood cyst?**
Weizhang Xiao, Jing Qin, Jia Feng, Feng Jiang, Ximming Chen, Xiang Cao, Qun Xue and Jiahai Shi

46 **Missile embolism from pulmonary vein to left ventricle: report of a case**
Mohammadrafie Khorgami, Fattaneh Khalaj, Maziar Gholampour and Hassan Tatari

51 **Case Report: Totally endoscopic minimally invasive mitral valve surgery during pregnancy: a case series**
Zhenzhong Wang, Lishan Zhong, Haijiang Guo, Yanli Liu, Chengbin Zhou, Yingxian Ye, Fengzhen Han and Huanlei Huang

56 **Retrosternal hematoma causing torsade de pointes after coronary artery bypass graft surgery: a case report**
Mohammadbagher Sharifkazemi, Mohammad Ghazinour, Mehrzad Lotfi, Soorena Khorshidi and Tahereh Davarpasand

63 **TMVR after TA-TAVR: a re-redo surgery—case report**
Nina Sophie Pommert, Thomas Puehler, Inga Voges, Stephanie Sellers and Georg Lutter

67 **Case Report: Left atrial dissection after mitral valve replacement: intraoperative management under TEE guidance**
Mengyan Wang, Fucheng Ji and Jinfeng Zhou

73 **Left ventricular outflow tract obstruction after transcatheter mitral valve replacement: a case report with a multifaceted approach**
Berenice Caneiro-Queija, Claudio E. Guerreiro, Julio Echarte-Morales, Rodrigo Estévez-Loureiro, Manuel Barreiro-Pérez, Rocío González-Ferreiro, Francisco Estévez-Cid, Juan José Legarra, Jose Antonio Baz and Andrés Íñiguez-Romo



OPEN ACCESS

EDITED AND REVIEWED BY

Hendrik Tevaearai Stahel,
University Hospital of Bern, Switzerland

*CORRESPONDENCE

Giuseppe Gatti
✉ gius.gatti@gmail.com

RECEIVED 14 November 2024

ACCEPTED 25 November 2024

PUBLISHED 09 December 2024

CITATION

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

COPYRIGHT

© 2024 Gatti. This is an open-access article distributed under the terms of the [Creative Commons Attribution License \(CC BY\)](#). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

Editorial: Case reports in heart surgery: 2023

Giuseppe Gatti*

Cardiac Surgery Unit, Cardio-Thoracic and Vascular Department, Azienda Sanitaria Universitaria Giuliano-Isontina (ASUGI), Trieste, Italy

KEYWORDS

myxoma, infective endocarditis, minimally invasive, multimodal imaging, transcatheter valve implantation, case report, heart surgery

Editorial on the Research Topic Case reports in heart surgery: 2023

I would like to state from the beginning that I am sincerely grateful to the Editors-in-Chief of *Frontiers in Cardiovascular Medicine* and *Frontiers in Surgery* for having asked me to coordinate the 2023 collection of "*Cases Reports in Heart Surgery*". I would also like to thank all the members of the Editorial Offices of the two valuable scientific Journals for having supported me at every moment of this experience. I hope I have been up to my task.

The objective of the present collection was to feature unique cases of patients that present with an unexpected diagnosis, treatment outcome, or clinical course. Only original Case Reports that significantly will advance the cardiac surgical field—at least in my opinion and that of the Reviewers who collaborated step by step with me in the manuscript review process—have been considered. Rare cases with typical features, frequent cases with atypical features, as well as cases with a convincing response to new treatments, were included in the present Research Topic.

The collection consists of 14 articles written by a total of 88 Authors from six countries (Canada, China, Germany, Iran, Spain and Switzerland) on three different continents. To date there have already been almost 13,000 total views.

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. For all these reasons, and to give clear objectives and more relevance to Case Reports sections, *Frontiers* has introduced these regular collections of original surgical cases. I think this editorial initiative is worthy and, personally, I am flattered by the invitation to coordinate it. Both for Heart Surgery and Interventional Cardiology, the most advanced frontiers of the disciplines are often glimpsed by analyzing Case Reports!

Very current issues are addressed in the present collection. These issues can be summarized as follows:

- The growing importance of minimally invasive surgery and interventional techniques and technologies [1, 3, 5, 14], and of their complications [1, 3, 14];
- The essential need to carefully plan the surgical strategy before operation [1, 6, 8, 12];

- The essential need of a multimodal imaging for complex lesions [1, 3, 6, 7, 8, 10, 12, 13];
- The need to develop specific surgical techniques for the treatment of infective endocarditis [11];
- The unusual presentations of “usual” lesions or complications following traditional heart surgery [2, 4, 7, 9, 10, 12, 13].

I synthesized the main message of each contribution to the present collection in **Table 1**.

To conclude, I would like to sincerely thank all the valuable Reviewers and Co-editors who helped me in my task. I have certainly learned a lot from them throughout this experience.

TABLE 1 Case reports in heart surgery 2023^a.

	First author	Title	Publication date	DOI	Total views at 14 November 2024	Main message	Key words
1.	Caneiro-Queija et al.	Left ventricular outflow tract obstruction after transcatheter mitral valve replacement: a case report with a multifaceted approach	Aug 21, 2024	doi.org/10.3389/fcvm.2024.1431639	856	Successful management using medical treatment and alcohol septal ablation of LVOT obstruction following TMVR. Authors discuss about this event and the strategies to prevent and manage the condition.	Alcohol septal ablation; LVOT obstruction; TMVR
2.	Wang et al.	Case Report: Left atrial dissection after mitral valve replacement: intraoperative management under TEE guidance	Aug 5, 2024	doi.org/10.3389/fcvm.2024.1413713	812	A successful TEE-guided surgical treatment of LA dissection following MV surgery. Indications and timing of surgery are discussed by the Authors.	LA dissection; MV replacement; TEE
3.	Pommert et al.	TMVR after TA-TAVR: a re-redo surgery—case report	May 20, 2024	doi.org/10.3389/fcvm.2024.1373840	1,032	A successful TMVR for mitral regurgitation early after transcatheter mitral valve repair in a patient undergone previously to two cardiac operations, TA-TAVR and CABG. The Authors emphasize the role of CT imaging in predicting both interaction between the prosthetic valves, and anatomy of the neo-LVOT.	Cardiac reoperation; CT imaging; LVOT; TA-TAVR; TMVR
4.	Sharifkazemi et al.	Retrosternal hematoma causing torsade de pointes after coronary artery bypass graft surgery; a case report	May 20, 2024	doi.org/10.3389/fcvm.2024.1331873	816	The Authors analyze a case of malignant arrhythmia refractory to medical treatment and due to a post-CABG retrosternal hematoma.	Arrhythmia; CABG; Retrosternal hematoma
5.	Wang et al.	Case Report: Totally endoscopic minimally invasive mitral valve surgery during pregnancy: a case series	Feb 26, 2024	doi.org/10.3389/fcvm.2024.1300508	1,445	A nice series of pregnant women with MV diseases were successfully treated using a totally endoscopic approach.	MV surgery; Minimally invasive surgery; Pregnancy;
6.	Khorgami et al.	Missile embolism from pulmonary vein to left ventricle: report of a case	Feb 23, 2024	doi.org/10.3389/fcvm.2024.1342146	969	Successful removal of a bullet, which was embolized from the pulmonary veins into the left ventricle of a child is reported.	Bullet; Embolism; Pediatric cardiac surgery
7.	Xiao et al.	Case Report: Giant left atrial cystic tumor: myxoma or intracardiac blood cyst?	Feb 14, 2024	doi.org/10.3389/fcvm.2024.1323890	1,816	Successful surgical removal of a rare case of giant LA cystic myxoma.	Cystic myxoma; Blood cyst; LA
8.	Buech et al.	Case Report: Incidental finding of an atresia of the inferior vena cava—a challenge for cardiac surgery	Feb 6, 2024	doi.org/10.3389/fcvm.2024.1321685	857	Perioperative management of IVC atresia during cardiopulmonary bypass. Preoperative diagnostics and intraoperative cannulation strategies to optimize venous drainage.	IVC atresia; Cannulation strategies; Cardiopulmonary bypass
9.	Tang et al.	Case Report: Acute cerebral infarction caused by left atrial and right ventricular myxoma received emergency operation	Jan 12, 2024	doi.org/10.3389/fcvm.2023.1316063	1,218	Insights from a rare case of ischemic stroke due to left atrial and right ventricular multiple cardiac myxoma.	Ischemic stroke; Multiple cardiac myxoma

(Continued)

TABLE 1 Continued

	First author	Title	Publication date	DOI	Total views at 14 November 2024	Main message	Key words
10.	Wu et al.	Case Report: Surgical management of idiopathic pulmonary aneurysms and review surgical approaches	Dec 20, 2023	doi.org/10.3389/fcvm.2023.1331982	1,885	Following the treatment of a patient with idiopathic pulmonary aneurysm, and by reviewing most of recent surgical strategies, the Authors have developed an original treatment flow chart for this challenging disease. The flowchart, which serves as a guide for the management of idiopathic pulmonary aneurysm, offers valuable insights and evidence-based recommendations. A special issue is the optimal approach for addressing the main pulmonary artery, its branches, and the pulmonary valve.	Flow chart; Idiopathic pulmonary aneurysm; Surgical strategies
11.	Chu et al.	Case Report: Pericardial patch repair of mitral annulus and mitral valve for a left atrial dissection caused by parasitic infective endocarditis	Nov 28, 2023	doi.org/10.3389/fcvm.2023.1239019	913	A complex mitral valve repair using a patch of autologous pericardium was performed to treat a rare case of left atrial dissection due to parasitic infectious endocarditis.	Autologous pericardium; MV repair; Parasitic infective endocarditis
12.	Jolou et al.	Case Report: Right atrial mass arising from the Eustachian valve	Nov 9, 2023	doi.org/10.3389/fcvm.2023.1268918	1,773	An organized thrombus attached to the Eustachian valve and Chiari network was found 18 months after a cardiac operation. After performing an interesting discussion on the possible etiology of the mass, the Authors emphasize the need, in this case, of a surgical re-exploration to perform diagnosis.	Cardiac thrombosis; Chiari network; Eustachian valve; Surgical re-exploration
13.	Zhou et al.	Case Report: Myocardial dissection caused by ruptured sinus of Valsalva aneurysm in association with a bicuspid aortic valve	Nov 8, 2023	doi.org/10.3389/fcvm.2023.1289624	1,139	The Authors present an interesting case of myocardial dissection due to rupture of a Valsalva sinus aneurysm. The presence of a bicuspid aortic valve may be a predisposing factor, as well as favor suspicion and early detection using multimodal imaging.	Bicuspid aortic valve; Multimodal imaging; Myocardial dissection; Sinus of Valsalva rupture
14	Wang et al.	Case report: pulmonary artery perforation during transseptal puncture for left atrial appendage closure requires emergency cardiac operation	Oct 10, 2023	doi.org/10.3389/fcvm.2023.1218582	1,769	The Authors review a case of successful rescue surgery for pulmonary artery perforation during percutaneous LA appendage closure. The causal mechanisms, clinical presentation and all possible management strategies are carefully discussed.	Percutaneous LA appendage closure; Pulmonary artery perforation; Rescue cardiac surgery

CABG, coronary artery bypass grafting; CT, computed tomography; DOI, digital object identifier; IVC, inferior vena cava; TA-TAVR, transapical transcatheter aortic valve replacement; TEE, transesophageal echocardiography; TMVR, transcatheter mitral valve replacement; LA, left atrial/atrium; LVOT, left ventricular outflow tract.

^aCase Reports are reported according to decrescent publication date.

Author contributions

GG: Writing – original draft, Writing – review & editing.

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.

Publisher's note

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.



OPEN ACCESS

EDITED BY

Giuseppe Gatti,
Azienda Sanitaria Universitaria Giuliano Isontina,
Italy

REVIEWED BY

José María Arribas,
Virgen de la Arrixaca University Hospital, Spain
Alfonso Agnino,
Humanitas Gavazzeni, Italy

*CORRESPONDENCE

Gang Ma
✉ sdbsmagang@163.com
Bo Li
✉ libosubmit@163.com

[†]These authors share first authorship

RECEIVED 09 May 2023

ACCEPTED 26 September 2023

PUBLISHED 10 October 2023

CITATION

Wang Y, Song B, Liu B, Zhang H, Bi C, Liu W, Ma G and Li B (2023) Case report: pulmonary artery perforation during transseptal puncture for left atrial appendage closure requires emergency cardiac operation. *Front. Cardiovasc. Med.* 10:1218582. doi: 10.3389/fcvm.2023.1218582

COPYRIGHT

© 2023 Wang, Song, Liu, Zhang, Bi, Liu, Ma and Li. This is an open-access article distributed under the terms of the [Creative Commons Attribution License \(CC BY\)](#). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

Case report: pulmonary artery perforation during transseptal puncture for left atrial appendage closure requires emergency cardiac operation

Yue Wang^{1,2†}, Beibei Song^{2†}, Bing Liu³, Hui Zhang², Chenglong Bi², Wenhao Liu^{2,4}, Gang Ma^{3*} and Bo Li^{2*}

¹Department of Clinical Medicine, Shandong First Medical University (Shandong Academy of Medical Sciences), Jinan, China, ²Department of Cardiology, Zibo Central Hospital, Zibo, China, ³Department of Cardiovascular Surgery, Zibo Central Hospital, Zibo, China, ⁴Zibo Central Hospital, Binzhou Medical University, Zibo, China

Patients with atrial fibrillation who take a high bleeding risk and are not candidates for oral anticoagulation therapy are increasingly being referred for left atrial appendage closure (LAAC) as an alternative method of stroke prevention. However, certain manipulations performed during the LAAC procedure, such as transseptal puncture (TSP), may potentially result in vessel injury and lead to cardiac tamponade or even fatality. Clinical significance and management strategies associated with these complications remain controversial. A 74-year-old female patient with atrial fibrillation was referred for left atrial appendage occlusion. During the puncture of the atrial septum, the catheter sheath inadvertently exited through the roof of the right atrium and continued to advance, resulting in pulmonary artery perforation. The patient underwent immediate pericardiocentesis and drainage, followed by surgical exploration for suturing the tear in the pulmonary artery and ligation of the left atrial appendage. This represents the first reported case of a pulmonary artery perforation occurring during a transseptal puncture procedure for left atrial appendage closure. The case exemplifies the feasibility of emergency cardiac surgery as a therapeutic intervention.

KEYWORDS

left atrial appendage closure, transseptal puncture, pulmonary artery perforation, cardiac tamponade, case report

Introduction

In recent years, advancements in electrophysiology and structural cardiac interventions have propelled the growing demand for transseptal procedures, primarily encompassing radiofrequency ablation, left atrial appendage closure, and mitral valve repair (1, 2). Left atrial appendage closure is currently an effective treatment for preventing ischemic stroke in patients with non-valvular atrial fibrillation and a high risk of bleeding and embolism, particularly those who cannot take oral anticoagulants due to contraindications or have a CHADS2 score of 2 or higher (3, 4). The manipulation during LAAC, however, has been associated with potentially serious complications including pericardial effusion, air embolization, device embolization, vascular lesions near the heart, ischemic stroke, and even death (5, 6). Physicians must possess a thorough comprehension of relevant complications and adeptly master their corresponding countermeasures. We present a

novel case of inadvertent pulmonary artery perforation during transseptal puncture in the context of proposed left atrial appendage occlusion, which was subsequently resolved through surgical suturing of the ruptured vessel and ligation of the left atrial appendage.

Case presentation

A 74-year-old female, was diagnosed with paroxysmal atrial fibrillation. Past medical history included coronary atherosclerotic heart disease, thrombosis in the left upper limb artery, heart failure, hypertension, ischemic cerebrovascular disease, and surgically treated endometrial cancer. Following coronary stent implantation and left upper extremity artery thrombectomy procedures, the patient received regular

treatment with standard-dose oral Aspirin, Clopidogrel sulfate, Pitavastine, Sacubitril-valsartan, Nifedipine, and Metoprolol. The patient presented with a CHA2DS2-VASC score of 7 and a HAS-BLED score of 3, clearly indicating the necessity for left atrial appendage closure. Considering the potential risks associated with long-term anticoagulation therapy, percutaneous LAAC is performed for secondary stroke prevention.

Preoperative left atrial CT angiography showed that the left atrial appendage was complex with a large branch, while no evident filling defect was observed in both the left atrial appendage and left atrium. No diverticulum or accessory atrial appendage was observed in the left atrium (Figure 1). Laboratory tests on the morning of surgery revealed a hemoglobin count of 117 g/L, a platelet count of $115 \times 10^9/L$, and an international normalized ratio (INR) of 0.96.

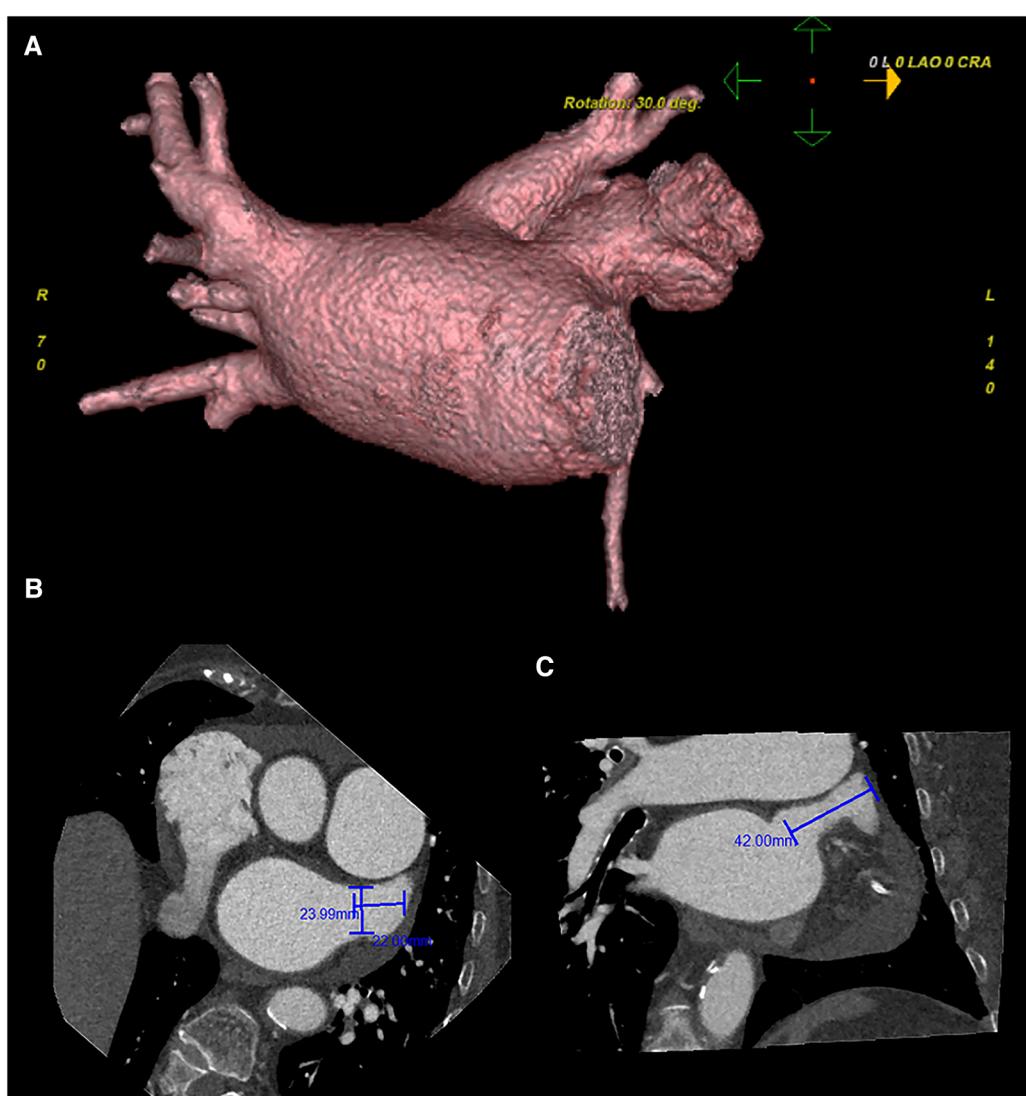


FIGURE 1

Preoperative cardiac CT angiography reveals a complex left atrial appendage with a large branch, no clear filling defect in the left atrial appendage and left atrium, and no diverticulum and accessory atrial appendage was identified in the left atrium (A) the cross-sectional image of the LAA orifice (B) the long diameter was about 2.4 cm, the short was about 2.2 cm. Oblique view (C) the maximum depth of the left atrial appendage branch was about 4.2 cm. Pericardial effusion.

The procedure was performed under digital subtraction angiography (DSA) guidance, with puncture of the right femoral vein, placement of a 6F sheath, and transseptal puncture using a catheter in the conventional manner. Afterwards, we promptly advanced the transseptal sheath and encountered difficulty in accessing the left superior pulmonary vein with the guidewire, while contrast injection indicated visualization of the pulmonary artery (Figure 2).

Patient experienced a sudden onset of hypotension and tachycardia, intraoperative echocardiography revealed the inadvertent insertion of the delivery tube sheath into the pulmonary artery, accompanied by a pericardial effusion measuring 17 mm. The ultrasound intervention department was promptly contacted for pericardiocentesis and drainage, while norepinephrine and dopamine were administered to augment blood pressure. Following active therapeutic interventions, hemodynamic stability was achieved in the patient.

Cardiac surgery team was consulted to perform surgical exploration in order to identify and suture the tear. A median sternotomy was performed under general anesthesia, and the pericardium was opened to locate the source of bleeding, revealing a subepicardial hematoma in the right atrioventricular groove. Exploration was initiated subsequent to the establishment of cardiopulmonary bypass, revealing that the delivery tube sheath had entered through the superior aspect of the right atrium and traversed through the posterior wall of the right pulmonary artery into the main pulmonary artery (Figure 3). The lacerations of the right atrium and pulmonary artery were repaired using 4-0 prolene sutures, followed by removal of the delivery tube sheath. Subsequently, the left atrial appendage was ligated with double 10-gauge sutures. However, there was still persistent bleeding from the pulmonary artery tear. The main pulmonary artery was dissected under cardiopulmonary bypass, revealing a right posterior wall tear upon exploration from within the pulmonary artery. The gasket from the pulmonary artery suture is reinforced with a 4-0

prolene line. The pulmonary artery incision is sutured with a 4-0 prolene line. Hemostasis is achieved by reinforcing the ruptured area of the pulmonary artery with a 4-0 prolene line. After thorough examination of the right atrial wall and pulmonary artery, the cardiopulmonary bypass machine was ceased upon confirming absence of any bleeding. Protamine was administered to neutralize the effects of heparin, followed by placement of pericardial and mediastinal drainage tubes post achieving hemostasis. The procedure was subsequently concluded in a conventional manner, with an intraoperative blood loss of approximately 600 ml.

Patient was transferred back to the intensive care unit for a day of close monitoring. On the second postoperative day, she returned to the cardiovascular surgery department and initiated anticoagulant therapy with low molecular weight heparin (LVMH). Two days later, she was transferred back to the cardiology department with a gradual reduction in pericardial and mediastinal drainage and discharged on postoperative day 25. Dual antiplatelet therapy with Indobufen and Clopidogrel bisulfate was regularly applied outside the hospital. After seven days, the patient returned to the outpatient clinic exhibiting no subjective symptoms and normalization of N-terminal pro-B-type natriuretic peptide levels (Table 1).

Discussion

Left atrial appendage closure has proven to be an effective alternative treatment to long-term oral anticoagulation for stroke prevention in patients with nonvalvular atrial fibrillation (3, 4). The advancement of interventional therapy relying on left heart access has propelled transseptal puncture to attain a highly standardized procedure (1, 2). Although transseptal puncture has an excellent safety profile, complications including cardiac perforation and tamponade, persistent atrial septal defect, thrombosis and embolism cannot be avoided (1, 2). Cardiac

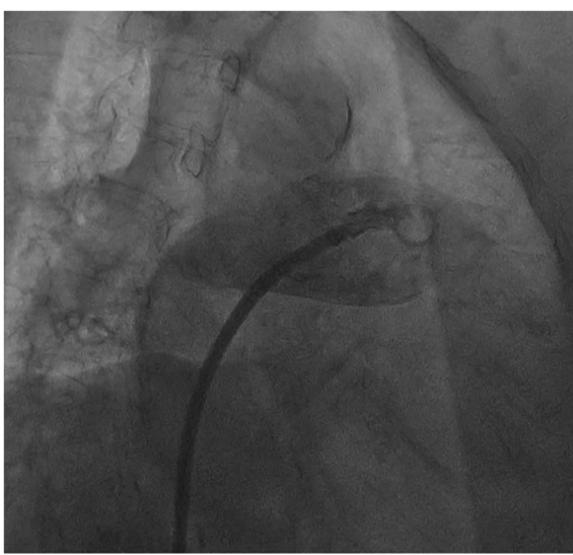


FIGURE 2
Angiography shows brisk contrast extravasation into the pulmonary artery space.

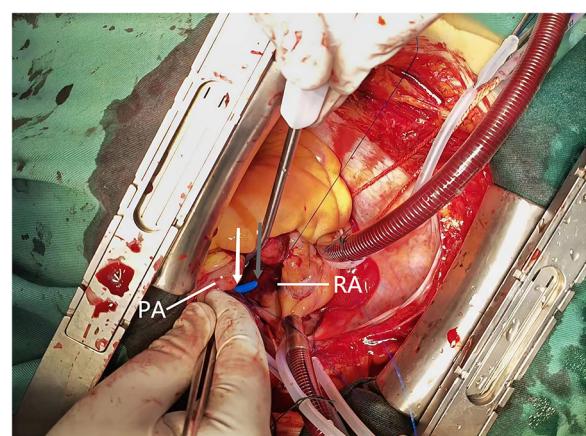


FIGURE 3
Intraoperative view reveals the pulmonary artery perforation caused by mispiercing of the delivery tube sheath. The grey arrows indicate the exit point of the delivery sheath from the roof of the right atrium, while the white arrows indicate its entry into the main pulmonary artery. PA, pulmonary artery; RA, right atrium.

TABLE 1 Timeline of events related to the patient's current procedure.

Time	Event
Baseline	Coronary atherosclerotic heart disease, thrombosis of the left upper limb artery, congestive heart failure, hypertension, ischemic cerebrovascular disease, endometrial cancer, contraindication for long-term anticoagulation.
Day 0	LAAC was performed under the guidance of DSA, angiography after atrial septal puncture showed pulmonary artery visualization. Urgent echocardiography revealed that the delivery tube sheath had inadvertently entered the pulmonary artery, triggering a pericardial effusion. Pericardiocentesis and drainage were performed and vasopressor drugs were given to maintain hemodynamic stability. Surgical exploration revealed the inadvertent puncture of the superior aspect of the right atrium by the birth sheath, which subsequently extended into the right posterior and main pulmonary arteries. The right atrium and pulmonary artery were repaired and sutured, while concurrently ligating the left atrial appendage. Pericardial and mediastinal drains were inserted for postoperative management.
Day 1	Treatment was continued in the ward, with the gradual reduction of pericardial effusion.
Day 25	Hospital discharge (Indobufen and Clopidogrel).
Day 32	Asymptomatic.

tamponade is a relatively common and highly lethal complication that can occur during transseptal puncture, with an overall incidence of approximately 1% (7). The underlying cause is perforation due to inadvertent advancement of the needle or sheath into the free atrial wall or adjacent vessels during transseptal puncture.

The operation of DSA-guided TSP is relatively straightforward and safe, but anatomical abnormalities may obscure the exact position of the needle or even mislead to the wrong puncture point. Therefore, it is crucial to evaluate the patient's left atrial appendage condition and anatomical morphology prior to surgery. Transesophageal echocardiography (TEE) enables clear determination of LAA thrombus presence, as well as measurement of basal diameter and depth, providing a foundation for the operator in selecting the appropriate occluder model. Cardiac CT angiography (CCTA) enables objective and accurate assessment of the left atrial appendage's shape, lobulation, and orifice parameters in the lateral aspect, providing a more comprehensive basis for determining the need for left atrial appendage occlusion. The utilization of these two methods enables the execution of multiplanar imaging and

three-dimensional reconstruction, thereby establishing a solid foundation for medical professionals to formulate treatment plans and assess potential risks. Additionally, cardiac magnetic resonance imaging (CMR) can also offer high-quality multiplanar images. For patients with contraindications or intolerance to TEE, intracardiac echocardiography (ICE) can serve as an alternative method for obtaining high-quality multiplanar images (8).

Previous cases have shown that pulmonary artery injury is the most common type of collateral vessel injury during left atrial appendage occlusion (9) which mostly occurs during LAA occluder implantation. The stabilization hook of ACP/Aamulet, the metal strut of Watchma (9, 10) or the LAmbe device (11) can result in pulmonary artery perforation. It often manifests as cardiac tamponade with hemodynamic failure, which can lead to death in severe cases (9). In the present case, we describe for the first time a pulmonary artery perforation that occurred during transseptal puncture. The mechanism of injury in this case may be that the atrial septal puncture point was too high, the delivery sheath accidentally penetrated the top of the right atrium and moved forward along the transverse pericardial sinus, leading to pulmonary artery perforation, which subsequently triggered cardiac tamponade (Figure 4).

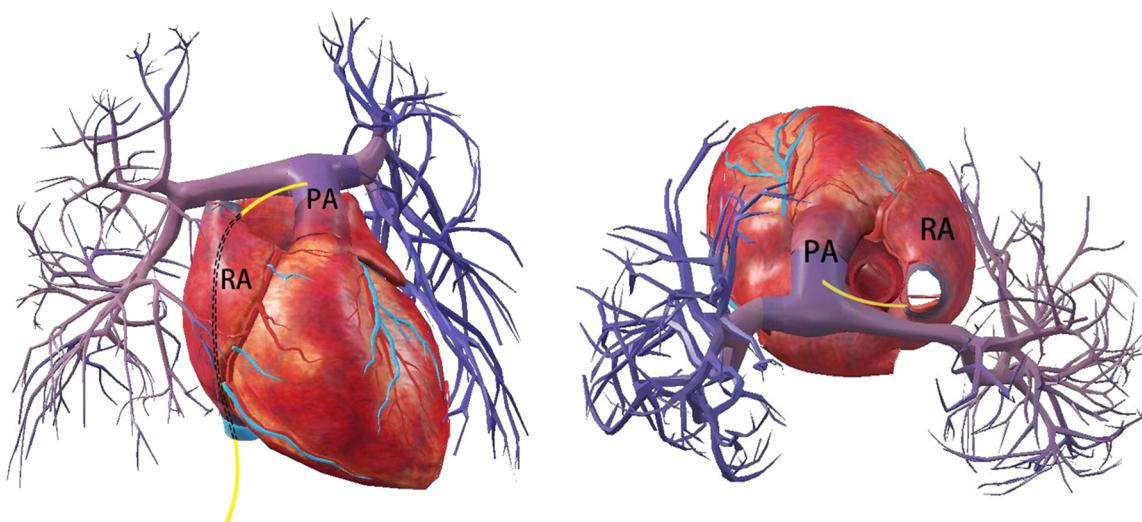


FIGURE 4

Schematic indicates that the delivery tube sheath enters the right atrium through the inferior vena cava, exits from the roof of the right atrium and punctures the main pulmonary artery leading to perforation.

Several cases have been reported in which a percutaneous occluder was utilized to block aortic or cardiac perforations (12, 7) during transseptal puncture. However, as cardiac tamponade can be life-threatening, emergency pericardiocentesis or surgical intervention appears to be the most promising therapeutic option to close the atrial leak (7). For hemodynamically unstable acute pericardial tamponade, ultrasound-guided pericardiocentesis and drainage should be performed first (1, 2). The accumulated blood can be withdrawn and observed if the bleeding volume is minimal and the bleeding rate is slow. The pigtail catheter can be inserted for continuous pericardial drainage in cases of significant and rapid bleeding. To avoid the fatal consequences of active bleeding, it is imperative to promptly request cardiac surgical intervention while ensuring continuous drainage. Fenestrated drainage and cardiac rupture sutures are crucial and effective treatments that should be performed urgently.

Combined with previous literature recommendations, when we encountered complications of pulmonary artery perforation, emergency cardiac decompression was performed immediately, followed by surgical suturing of the ruptured artery and ligation of the left atrial appendage in order to avoid active bleeding. Postoperative follow-up showed that the patient recovered well, which confirmed that surgical repair was an effective treatment.

Conclusion

Pulmonary artery perforation is a rare but critical complication associated with left atrial appendage closure, which may occur during transseptal puncture. Clinicians should maintain vigilance for uncommon complications and actively enhance preoperative assessment in order to decrease the occurrence of complications. In the event of such complications, emergency surgical exploration and ligation of the left atrial appendage represent a feasible treatment option.

Patient perspective

“Although a rare complication occurred and the left atrial appendage closure was not successfully completed, I was fortunate to undergo timely surgical intervention, ensuring my safety. I express my gratitude to the medical staff for their exceptional treatment. I hope that my experience can serve as a valuable reference for similar cases”.

References

1. O'Brien B, Zafar H, De Freitas S, Sharif F. Transseptal puncture—review of anatomy, techniques, complications and challenges. *Int J Cardiol.* (2017) 233:12–22. doi: 10.1016/j.ijcard.2017.02.009
2. Alkhouri M, Rihal CS, Holmes DR Jr. Transseptal techniques for emerging structural heart interventions. *JACC Cardiovasc Interv.* (2016) 9(24):2465–80. doi: 10.1016/j.jcin.2016.10.035
3. Holmes DR, Reddy VY, Turi ZG, Doshi SK, Sievert H, Buchbinder M, et al. Percutaneous closure of the left atrial appendage versus warfarin therapy for prevention of stroke in patients with atrial fibrillation: a randomised non-inferiority trial. *Lancet.* (2009) 374(9689):534–42. doi: 10.1016/S0140-6736(09)61343-X Erratum in: Lancet. 2009 Nov 7;374(9701):1596.
4. Hindricks G, Potpara T, Dagres N, Arbelo E, Bax JJ, Blomström-Lundqvist C, et al. 2020 ESC guidelines for the diagnosis and management of atrial fibrillation developed in collaboration with the European association for cardio-thoracic surgery (EACTS): the task force for the diagnosis and management of atrial fibrillation of the European society of cardiology (ESC) developed with the special contribution of the European heart rhythm association (EHRA) of the ESC. *Eur Heart J.* (2021) 42(5):373–498. doi: 10.1093/eurheartj/ehaa612 Erratum in: Eur Heart J. 2021 Feb 1;42(5):507. Erratum in: Eur Heart J. 2021 Feb 1;42(5):546–547. Erratum in: Eur Heart J. 2021 Oct 21;42(40):4194.

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

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

Author contributions

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

Funding

This study was funded by two grants from the Natural Science Foundation of Shandong Province (Grant Number: ZR2020MH025 and ZR2023MH136).

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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

5. Reddy VY, Doshi SK, Sievert H, Buchbinder M, Neuzil P, Huber K, et al. Percutaneous left atrial appendage closure for stroke prophylaxis in patients with atrial fibrillation: 2.3-year follow-up of the PROTECT AF (watchman left atrial appendage system for embolic protection in patients with atrial fibrillation) trial. *Circulation*. (2013) 127(6):720–9. doi: 10.1161/CIRCULATIONAHA.112.114389
6. Boersma LV, Schmidt B, Betts TR, Sievert H, Tamburino C, Teiger E, et al. Implant success and safety of left atrial appendage closure with the WATCHMAN device: peri-procedural outcomes from the EWOLUTION registry. *Eur Heart J*. (2016) 37(31):2465–74. doi: 10.1093/euroheartj/ehv730
7. Schamroth Pravda N, Codner P, Vaknin Assa H, Hirsch R. Management of ascending aorta perforation during transseptal puncture for left atrial appendage closure: a case report. *Eur Heart J Case Rep.* (2021) 5(4):ytab154. doi: 10.1093/euroheartj/ytab154
8. Laterra G, Dattilo G, Correale M, Brunetti ND, Artale C, Sacchetta G, et al. Imaging modality to guide left atrial appendage closure: current status and future perspectives. *J Clin Med.* (2023) 12(11):3756. doi: 10.3390/jcm12113756
9. Sharma SP, Murtaza G, Madoukh B, Atkins D, Nydegger C, Jeffery C, et al. Systematic review of contiguous vessel and valve injury associated with endocardial left atrial appendage occlusion devices. *J Atr Fibrillation*. (2019) 12(2):2256. doi: 10.4022/jafib.2256
10. Lu C, Zeng J, Meng Q, Zeng Z. Pulmonary artery perforation caused by a left atrial appendage closure device. *Catheter Cardiovasc Interv.* (2020) 96(7):E744–6. doi: 10.1002/ccd.28541
11. Bretones-Pino T, Rivera-López R, Carrero-Castaño A, Molina-Navarro E. Pulmonary artery perforation by atrial appendage closure device and the contribution of auricular contraction to its pathogenesis. *CJC Open.* (2021) 4(1):97–9. doi: 10.1016/j.cjco.2021.07.021
12. Bai Y, Zhang S, Qin YW, Zhao XX. Percutaneous occlusion of transseptal puncture-related free wall perforation at the coronary sinus with a ventricular septal occluder during left atrial appendage closure: a case report. *Catheter Cardiovasc Interv.* (2020) 96(7):E755–7. doi: 10.1002/ccd.28589
13. Seppelt P, Karck M, Kallenbach K. Aortic mispuncture during routine catheterization requires emergency cardiac operation. *Thorac Cardiovasc Surg.* (2013) 61(7):610–11. doi: 10.1055/s-0033-1333846



OPEN ACCESS

EDITED BY

Giuseppe Gatti,
Azienda Sanitaria Universitaria Giuliano Isontina,
Italy

REVIEWED BY

Thierry Caus,
University of Picardie Jules Verne, France
Bardia Arbabhani,
Leiden University Medical Center (LUMC),
Netherlands

*CORRESPONDENCE

Chengde Liao
✉ chengdeliao@qq.com
Yu Jiang
✉ 77913403@163.com

RECEIVED 11 September 2023

ACCEPTED 26 October 2023

PUBLISHED 08 November 2023

CITATION

Zhou X, Xu Y, He Q, Tan N, Chu J, Liu B, Zhu Y, Liao C and Jiang Y (2023) Case Report: Myocardial dissection caused by ruptured sinus of Valsalva aneurysm in association with a bicuspid aortic valve. *Front. Cardiovasc. Med.* 10:1289624. doi: 10.3389/fcvm.2023.1289624

COPYRIGHT

© 2023 Zhou, Xu, He, Tan, Chu, Liu, Zhu, Liao and Jiang. This is an open-access article distributed under the terms of the [Creative Commons Attribution License \(CC BY\)](#). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

Case Report: Myocardial dissection caused by ruptured sinus of Valsalva aneurysm in association with a bicuspid aortic valve

Xinyan Zhou¹, Yan Xu¹, Qian He², Na Tan¹, Jixiang Chu³, Bin Liu¹, Yu Zhu³, Chengde Liao^{1*} and Yu Jiang^{4*}

¹Department of Radiology, Kunming Yan'an Hospital (Yan'an Hospital Affiliated to Kunming Medical University), Kunming, China, ²Department of Ultrasound, Kunming Yan'an Hospital (Yan'an Hospital Affiliated to Kunming Medical University), Kunming, China, ³Department of Radiology, Yunnan Cancer Hospital (The Third Affiliated Hospital of Kunming Medical University), Kunming, China, ⁴Department of Cardiovascular Surgery, Kunming Yan'an Hospital (Yan'an Hospital Affiliated to Kunming Medical University), Kunming, China

In this report, we present a case of left-right sinus fusion in a Ruptured sinus of Valsalva aneurysm (RSVA) that perforated into the myocardium, giving rise to myocardial dissection. The existence of an anomalous bicuspid aortic valve (BAV) is contemplated as a potential etiological element in this context. Employing multimodal imaging modalities, encompassing transthoracic echocardiography and computed tomography (CT), facilitated the visualization of a dissecting hematoma situated within the myocardium subsequent to the RSVA. Following this, our patient underwent an Cabrol surgical intervention, received patch repair, and underwent mitral valve annuloplasty, during which a three-year period transpired without the occurrence of any deleterious cardiac events. In summary, this report establishes the cornerstone for the surgical intervention of RSVA, shedding light on the efficacious handling of RSVA-associated myocardial dissection. It posits that the presence of a BAV may serve as a predisposing factor to RSVA rupture, potentially elevating the susceptibility to myocardial dissection. The utilization of diverse multimodal imaging methodologies played an indispensable role in the detection of a hematoma within the myocardial tissue subsequent to the RSVA rupture. The uneventful three-year postoperative follow-up of the patient underscores the efficacy of the undertaken interventions.

KEYWORDS

sinus of Valsalva aneurysm, bicuspid aortic valve, myocardial dissection, echocardiogram, computed tomography

1. Introduction

Sinus of Valsalva aneurysm (SVA) is a rare cardiac anomaly, with an incidence of approximately 0.09% (1). This is primarily attributed to structural defects existing between the aortic media and fibrous ring (2). RSVA is an uncommon complication of SVA that may rupture into the cardiac chamber or the pericardial cavity, but seldom involves myocardial tearing. Patients with a bicuspid aortic valve, characterized by a two-leaflet aortic valve anomaly, often exhibit an increased risk of ascending aortic dilation due to alterations in blood flow patterns. Existing literature suggests (3) a connection between the BAV and the development of SVA. This implies that BAV may serve as a risk factor for RSVA and could potentially be associated with the interventricular septal

and myocardial intramural hematoma following SVA rupture. This case presents an instance of BAV anomaly and RSVA, resulting in an interventricular and intramural myocardial hematoma.

2. Case report

A 60-year-old Chinese male presented with upper abdominal pain accompanied by dizziness and palpitations for 2 days. The patient's heart function was classified as NYHA functional class IV, and vital signs and blood biochemistry tests showed no critical indicators. Transthoracic echocardiography (TTE) revealed a myocardial dissection with a pouch-like protrusion into the right ventricle, interrupted wall echo in the septum and the basal segment of the left ventricular anterior wall, with an adherent hyperechoic structure within (Figure 1). A dual-source CT scan shows the diameter of the ascending aorta to be approximately 4.8 cm. There was a BAV and fusion of the left and right aortic sinuses, with an outward protrusion forming a cavity in the anterior aortic sinus. Myocardial dissection formation, likely due to rupture of the anterior aortic sinus, was suspected. The neck of the aneurysm had a diameter of

approximately 1.4 cm, and the morphology of the aneurysm was irregular, with localized pushing on the left coronary artery (Figure 2).

After multidisciplinary consultation, the patient was diagnosed with a bicuspid aortic valve anomaly combined with a ruptured sinus of Valsalva and intramural hematoma within the interventricular septum, the basal segment of the left ventricular anterior wall, and the anterior lateral wall. Because of the patient's preexisting valve condition, we also noted the presence of moderate mitral valve regurgitation and tricuspid valve regurgitation. During our surgical exploration, we observed the formation of a partial thrombus within the heart.

After obtaining the patient's informed consent, surgical intervention was initiated. The procedure commenced with a midline sternotomy, revealing a dilated ascending aortic aneurysm upon opening the pericardium. Notable flutter was observed in the pulmonary artery and the conus of the right ventricle. Aortic and bicaval cannulation was performed to initiate extracorporeal circulation. Intraoperative exploration revealed left ventricular enlargement, bicuspid aortic valve deformity with anterior-posterior orientation, leaflet thickening, and calcification. A longitudinal tear of

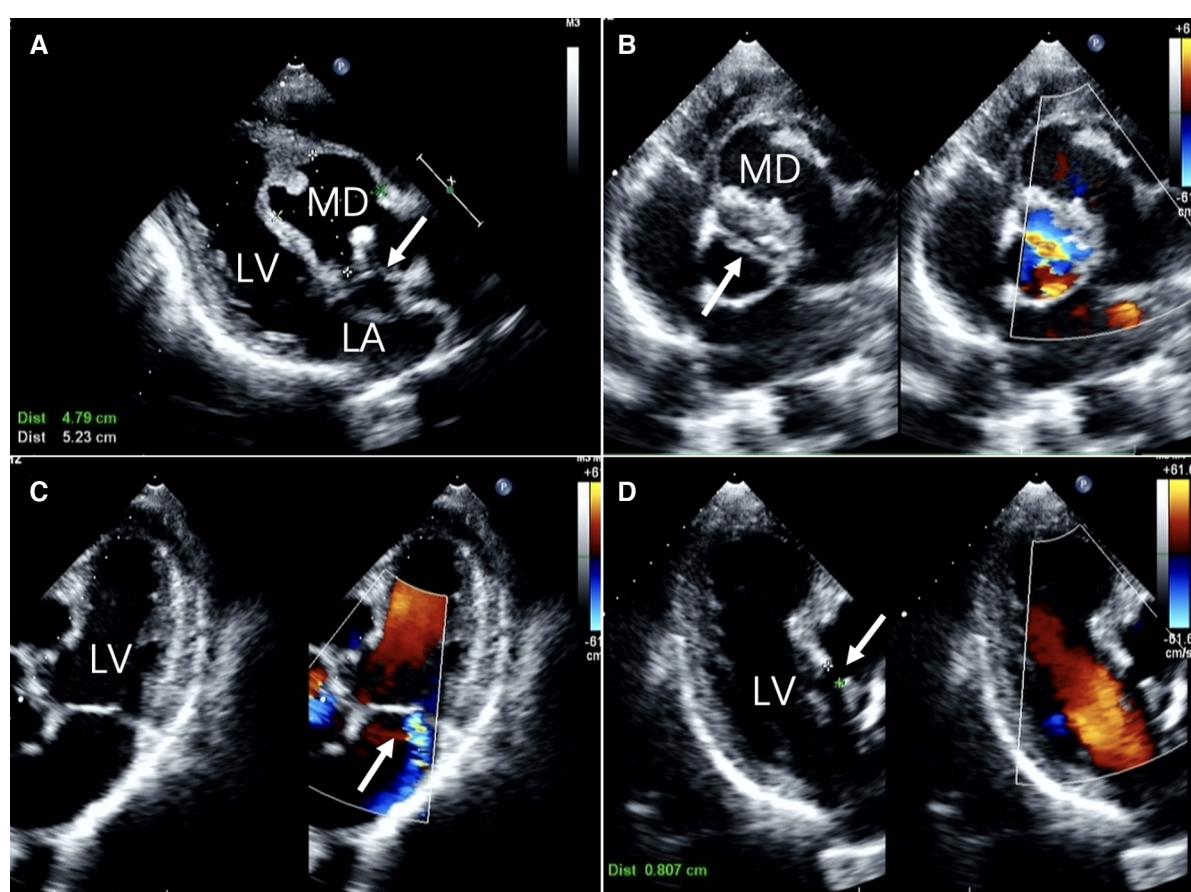


FIGURE 1

Two-dimensional transthoracic echocardiographic images (A) long-axis view of the left ventricle showing echo interruption, no echo in the interventricular septum. (white arrow) (B) the short-axis cross-sectional view of the aorta demonstrates a bicuspid aortic valve anomaly. (white arrow) (C) the non-standard pentachamber cardiac section reveals mitral valve eccentric regurgitation. (D) The three-chamber cardiac cross-section reveals the dimension of the orifice. LA, left atrium; LV, left ventricle; MD, Myocardial dissection.

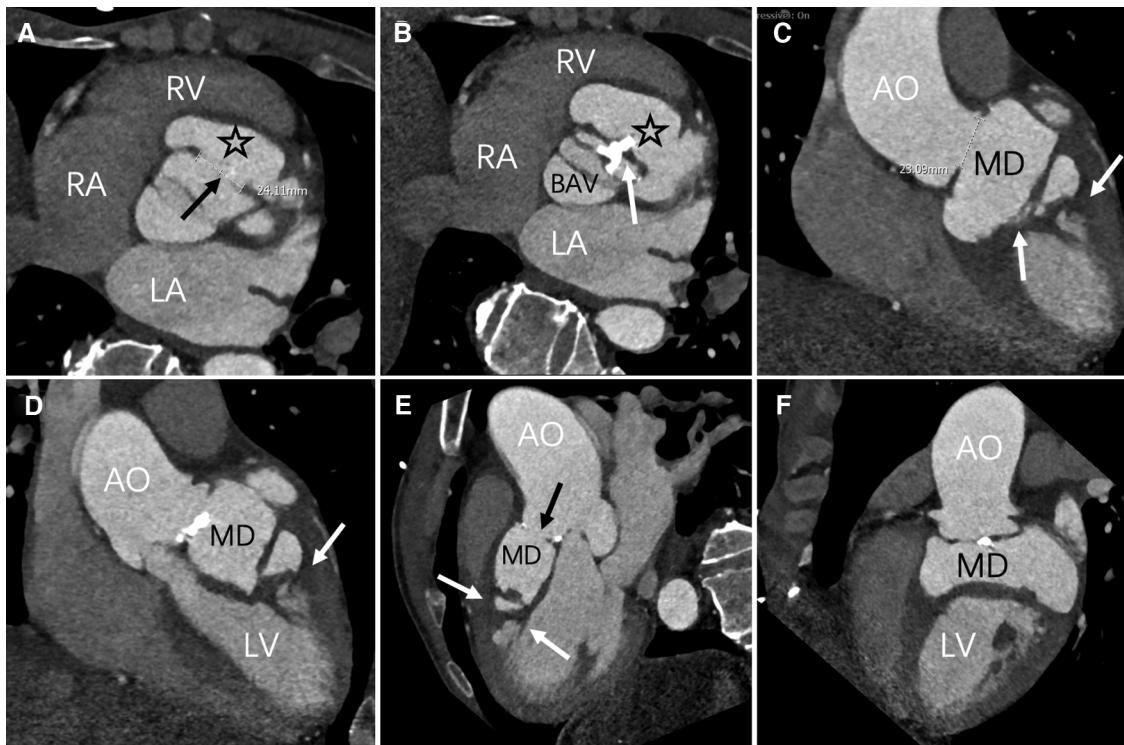


FIGURE 2

Axial position, contrast-enhanced CT imaging reveals the maximum aperture of the sinus of Valsalva aneurysm to be 2.4 cm (black arrow). The myocardial dissection originates from the fused left and right coronary sinuses (asterisk). A bicuspid aortic valve anomaly is evident, with valve leaflet thickening and calcification (white arrow) (C–F) multiPlanar Reconstruction (MPR) exhibits irregular MD, fusion of the left and right coronary sinuses (black arrow), and high-density opacities within the ventricular wall and interventricular septum (white arrow). LA, left atrium; LV, left ventricle; RA, right atrium; RV, right ventricle; AO, Aortic; MD, Myocardial dissection; BAV, bicuspid aortic valve.

approximately 2 cm was evident in the aortic sinus, extending all the way to the interventricular septum and the posterior wall of the left ventricle, accompanied by the formation of an intracavitary thrombus (Figure 3). Following the exploration, repairs of the aortic sinus, the Cabrol procedure, and annuloplasty of the aortic valve were performed. We utilized a 21 mm mechanical valve prosthesis for the aortic valve replacement and completed interrupted sutures along the aortic annulus. Subsequently, aortic replacement was carried out using dacron tube, and approximately 0.7 cm of prosthetic material was employed for indirect coronary reimplantation in the Cabrol procedure. Subsequent transesophageal echocardiography revealed no significant regurgitation, and myocardial function was within the normal range. Closure of the atrial septum and the right atrium was performed. Partial pericardial closure was meticulously carried out to ensure hemostasis, followed by chest closure.

Postoperatively, the patient experienced a smooth recovery, with normal mechanical valve opening and closing sounds. CT scans revealed a satisfactory anastomosis of the aorta and coronary arteries, while transthoracic echocardiography (TTE) indicated a significant improvement in left ventricular ejection fraction and anterior wall motion (Figure 4). During a three-year follow-up, the patient's outcome remained favorable.

3. Discussion

SVA is an uncommon cardiac anomaly. According to a series of postmortem examinations (1), the incidence rate of SVA is recorded at 0.09%, with a gender ratio of 3:1 (4, 5). SVA predominantly manifests among individuals aged 30 to 45, with a heightened prevalence among individuals of Asian descent (6). The embryonic basis of SVA is linked to impaired development of the far basal part of the membranous interventricular septum. It often involves the right coronary sinus, followed by the contiguous two-thirds of the non-coronary sinus. In instances of SVA rupture, the most common occurrence is rupture into the right ventricle, followed by the right atrium (7). There exists a mere 2% likelihood of penetration into the interventricular septum and the ventricular myocardium (5). In this specific case, an Asian patient experienced SVA rupture into the interventricular septum and the left ventricular myocardium due to the fusion of the left and right coronary sinuses caused by BAV.

This International evidence-based nomenclature on the congenital bicuspid aortic valve and its aortopathy recognizes Fused type, 2-sinus type with 2 phenotypes and Partial-fusion or forme fruste (8). In this particular case, the patient presented with L-R type. BAV is associated with genetic mutations involving GATA5, NOTCH1, ACTA2, and others (9). Patients with BAV may experience fusion of the aortic sinuses, resulting in altered blood

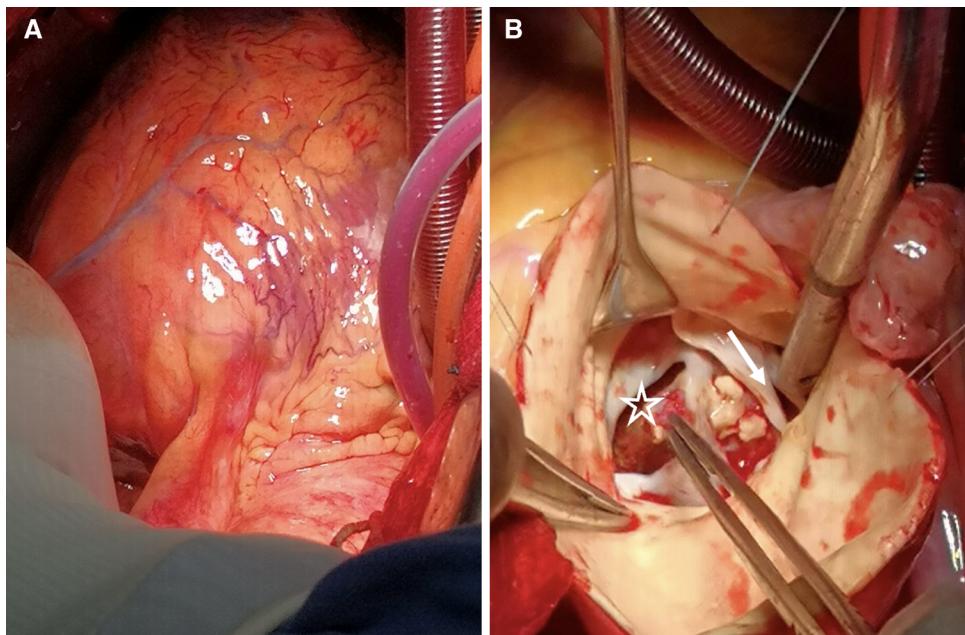


FIGURE 3

(A) The heart's presentation reveals evident edema. (B) BAV (white arrow) is discernible, accompanied by numerous calcifications on the valve leaflets. Additionally, we observe the occurrence of a sinus aneurysm rupture (asterisk), concomitant with an intraluminal thrombus.

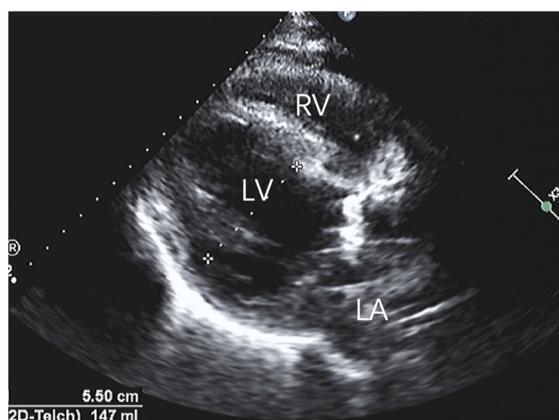


FIGURE 4

After repairing the rupture of the aortic sinus, the myocardial dissection resolved, and the anatomical structure returned to its normal state.

flow patterns that often lead to vascular wall stress overload. This, in turn, can lead to the loss of vascular smooth muscle cells, reduced fibrous protein content, elastic fiber rupture, and matrix disruption (10). Consequently, it could potentially increase the risk of SVA rupture and exacerbate the extent of involvement following SVA rupture. Our patient is afflicted with BAV, which renders the aortic wall vulnerable. When the SVA abruptly ruptures, the force of blood flow creates openings in the vessels and gives rise to interventricular septal defects and blood shunting. As blood continues to impact the vessel walls and endocardium, it gradually forms an intramural hematoma between the interventricular septum and the ventricular myocardium.

In a recent comprehensive review, it was determined that approximately 50% of RSVA patients manifest respiratory distress, followed by chest pain (18%). Symptoms such as palpitations, syncope, vomiting, and fever were also noted (3, 11). In this particular case, the patient presented solely with symptoms of chest pain. Interestingly, nearly 10% of patients remain asymptomatic (11). Without prompt intervention, nearly 80% of cases may progress to heart failure and sudden death, leading to a bleak prognosis for affected individuals. Hence, accurate diagnosis and timely treatment are of paramount significance.

In general, echocardiography demonstrates exceptional sensitivity to both vascular walls and myocardium. Within the ultrasound examination, our patient's presentation was characterized by an aortic sinus that protruded into the right ventricle, displaying an interrupted wall echo. Notably, there is an anomalous echo-free region in the basal segment of the interventricular septum and the left ventricular anterior wall. This manifestation signifies the rupture of the sinus aneurysm, resulting in the tearing of the corresponding interventricular septum and myocardium.

CT imaging revealed a bicuspid valve in an L-R cusp fusion in this patient, with multiple calcifications on the leaflets. The merging of the left and right coronary sinuses forms a pouch-like structure. An irregularly shaped high-density shadow appears on the interventricular septum and the left ventricular wall, indicative of a myocardial dissection within the myocardium. CT provides high-resolution three-dimensional images, which offer a more intuitive basis for clinical decision-making and complement the results of echocardiography (12).

The surgical treatment of RSVA was first reported by Lillehei and colleagues in 1957 (13). Currently, treatment is typically

guided by international guidelines (14), following a similar approach as in patients with aortic root tumors. In our case, both CT scans and surgical exploration revealed myocardial dissection in the interventricular septum and the anterior wall of the left ventricle. To address these related defects and excise the aneurysm sac, thereby preventing cardiac obstruction or aortic valve dysfunction, we chose to employ bovine pericardial patch repair of the aortic sinus rupture. The literature suggests (15) that aortic valve replacement and ascending aortic replacement may provide stability to the sinus in patients with bicuspid valve anomalies and aortic root dilation. Consequently, we performed an aortic valve replacement and carried out the Cabrol procedure. Due to structural damage to the aorta, we chose the Cabrol procedure instead of the Bentall procedure.

In summary, RSVA patients with concomitant BAV often exhibit valve leaflet anomalies, necessitating meticulous exploration and complex surgical approaches for personalized repair and treatment.

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 Medical Ethical committee of Yan'an Affiliated Hos. 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. Written informed consent was obtained from the participant/patient(s) for the publication of this case report.

Author contributions

XZ: Writing – original draft, Conceptualization, Data curation. YX: Data curation, Writing – original draft. QH: Writing – review & editing, Investigation, Software, Writing – original draft. NT:

Formal Analysis, Writing – original draft. JC: Data curation, Writing – original draft. BL: Writing – review & editing. YZ: Writing – review & editing. CL: Conceptualization, Funding acquisition, Writing – review & editing. YJ: Conceptualization, 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 Yunnan Talents Support Program [grant number XDYC-MY-2022-0064].

Acknowledgments

The success of this study is attributed to the hard work and support of many individuals. We want to express our gratitude to each and every person who contributed to this research; their assistance has been truly invaluable.

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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

Supplementary material

The Supplementary Material for this article can be found online at: <https://www.frontiersin.org/articles/10.3389/fcvm.2023.1289624/full#supplementary-material>

References

1. Weinreich M, Yu PJ, Trost B. Sinus of Valsalva aneurysms: review of the literature and an update on management. *Clin Cardiol.* (2015) 38(3):185–9. doi: 10.1002/clc.22359
2. Edwards JE, Burchell HB. The pathological anatomy of deficiencies between the aortic root and the heart, including aortic Sinus aneurysms. *Thorax.* (1957) 12(2):125–39. doi: 10.1136/thx.12.2.125
3. Moustafa S, Mookadam F, Cooper L, Adam G, Zehr K, Stulak J, et al. Sinus of Valsalva aneurysms—47 years of a single center experience and systematic overview of published reports. *Am J Cardiol.* (2007) 99(8):1159–64. doi: 10.1016/j.amjcard.2006.11.047
4. Takach TJ, Reul GJ, Duncan JM, Cooley DA, Livesay JJ, Ott DA, et al. Sinus of Valsalva aneurysm or fistula: management and outcome. *Ann Thorac Surg.* (1999) 68(5):1573–7. doi: 10.1016/S0003-4975(99)01045-0

5. Bricker AO, Ayutu B, Mohammed TL, Williamson EE, Syed IS, Julsrud PR, et al. Valsalva sinus aneurysms: findings at CT and MR imaging. *RadioGraphics*. (2010) 30(1):99–110. doi: 10.1148/rg.301095719
6. Sarikaya S, Adademir T, Elibol A, Büyükbayrak F, Onk A, Kirali K. Surgery for ruptured sinus of Valsalva aneurysm: 25-year experience with 55 patients. *Eur J Cardiothorac Surg.* (2013) 43(3):591–6. doi: 10.1093/ejcts/ezs450
7. Feldman DN, Roman MJ. Aneurysms of the sinuses of Valsalva. *Cardiology*. (2006) 106(2):73–81. doi: 10.1159/000092635
8. Michelena H I, Della Corte A, Evangelista A, Maleszewski JJ, Edwards WD, Roman MJ, et al. International consensus statement on Nomenclature and classification of the congenital bicuspid aortic valve and its aortopathy, for clinical, surgical, interventional and research purposes. *Ann Thorac Surg.* (2021) 112(3):e203–e35. doi: 10.1016/j.athoracsur.2020.08.119
9. Presti L, Guzzardi DG F, Bancone C, Fedak PWM, Della Corte A. The science of BAV aortopathy. *Prog Cardiovasc Dis.* (2020) 63(4):465–74. doi: 10.1016/j.pcad.2020.06.009
10. Wilton E, Jahangiri M. Post-stenotic aortic dilatation. *J Cardiothorac Surg.* (2006) 1(1):7. doi: 10.1186/1749-8090-1-7
11. Marroush TS, Boshara AR, Botros B, Venditti P, Ahmed Z, Dawood L, et al. Rupture of sinus of Valsalva aneurysm: two case reports and a concise review of the literature. *Heart Lung*. (2018) 47(2):131–5. doi: 10.1016/j.hrtlng.2017.10.005
12. Ikonomidis I, Makavos G, Katsimbri P, Boumpas DT, Parissis J, Iliodromitis E. Imaging risk in multisystem inflammatory diseases. *JACC: Cardiovascular Imaging*. (2019) 12(12):2517–37. doi: 10.1016/j.jcmg.2018.06.033
13. Lillehei CW, Stanley P, Varco RL. Surgical treatment of ruptured aneurysms of the sinus of Valsalva.
14. 2014 ESC guidelines on the diagnosis and treatment of aortic diseases: document covering acute and chronic aortic diseases of the thoracic and abdominal aorta of the adultThe task force for the diagnosis and treatment of aortic diseases of the European society of cardiology (ESC). *Eur Heart J.* (2014) 35(41): 2873–926. doi: 10.1093/eurheartj/ehu281
15. Milewski RK, Habertheuer A, Bavaria JE, Siki M, Szeto WY, Krause F, et al. Fate of remnant sinuses of Valsalva in patients with bicuspid and trileaflet valves undergoing aortic valve, ascending aorta, and aortic arch replacement. *J Thorac Cardiovasc Surg.* (2017) 154(2):421–32. doi: 10.1016/j.jtcvs.2017.03.150



OPEN ACCESS

EDITED BY

Giuseppe Gatti,
Azienda Sanitaria Universitaria Giuliano
Isontina, Italy

REVIEWED BY

James Horowitz,
New York University, United States
Ilaria Radano,
Hospital Mauritian Turin, Italy

*CORRESPONDENCE

Christoph Huber
✉ christoph.huber@hcuge.ch

RECEIVED 28 July 2023

ACCEPTED 23 October 2023

PUBLISHED 09 November 2023

CITATION

Jolou J, Martineau J, Müller H, Cikirkcioglu M and Huber C (2023) Case Report: Right atrial mass arising from the Eustachian valve. *Front. Cardiovasc. Med.* 10:1268918. doi: 10.3389/fcvm.2023.1268918

COPYRIGHT

© 2023 Jolou, Martineau, Müller, Cikirkcioglu and Huber. This is an open-access article distributed under the terms of the [Creative Commons Attribution License \(CC BY\)](#). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

Case Report: Right atrial mass arising from the Eustachian valve

Jalal Jolou¹, Jérôme Martineau¹, Hajo Müller²,
Mustafa Cikirkcioglu¹ and Christoph Huber^{1*}

¹Department of Cardiovascular Surgery, Geneva University Hospitals, Geneva University, Geneva, Switzerland, ²Department of Cardiology, Geneva University Hospitals, Geneva University, Geneva, Switzerland

A mass in the right atrium (RA) is an unusual finding that warrants further investigation. We report the case of a 72-year-old male patient who underwent a Bentall operation with a biological composite graft and closure of patent foramen ovale 18 months prior to his presentation with an incidental new RA mass during follow-up echocardiography. Transesophageal echocardiography and thoracic CT angiography confirmed a right atrial mass attached to the Eustachian valve and additionally revealed a non-occlusive pulmonary embolism in the inferior lobar artery of the left lung. Despite 2 months of anticoagulation treatment, the size of the mass did not decrease. Further MRI imaging showed a central mass enhancement which raised concerns about a tumoral lesion. Following a discussion with the local Heart Team, management with surgical treatment was decided. The intraoperative findings revealed a 2.5 cm × 2.1 cm mass arising from the Eustachian valve and a non-diagnosed Chiari network in the RA. Both were resected and sent for a frozen section procedure which excluded a malignancy. The final histopathological analysis described fibrotic tissues compatible with an organized thrombus. The patient was discharged on postoperative day 7 without any complications. Although imaging studies are useful for the initial and differential diagnosis of RA masses, it is not always possible to get the final diagnosis without surgery. In case of a suspicion of a potentially malignant pathology, surgical exploration and resection are necessary.

KEYWORDS

Eustachian valve, right atrial mass, Chiari network, intra-cardiac mass, thrombi, cardiac MRI

Introduction

A mass in the right atrium (RA) is an unusual finding that deserves further investigation. Nowadays, their incidence has increased due to more frequent imaging studies. We report the case of a 72-year-old male patient who had a right atrial mass in a very unusual localization on the Eustachian valve of the RA.

Case description

A 72-year-old male patient who had undergone a Bentall operation with a biological composite graft and closure of a patent foramen ovale 18 months before, presented with an incidental new RA mass during a follow-up echocardiography. Transesophageal echocardiography and thoracic CT angiography confirmed the presence of the right atrial mass and its location on the Eustachian valve (**Figures 1A,B**). Moreover, an incidental non-occlusive pulmonary embolism in the left inferior lobe pulmonary artery was diagnosed (**Figure 1C**). Despite 2 months of oral anticoagulant treatment, the size of the

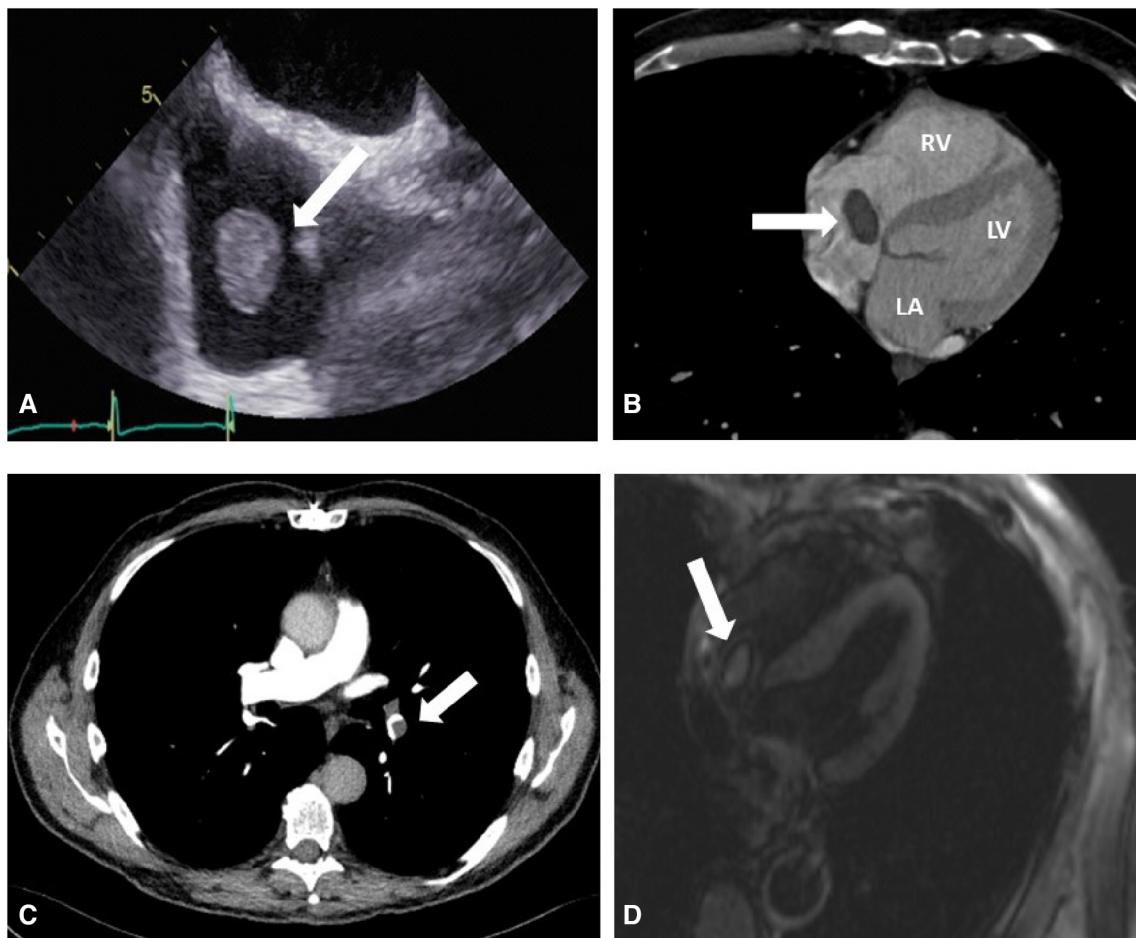


FIGURE 1

(A) Preoperative transesophageal echocardiography shows a mobile right atrial mass (arrow) on the Eustachian valve. (B) Preoperative thoracic CT angiography showing the right atrial mass (arrow), right ventricle (RV), left atrium (LA), and left ventricle (LV). (C) Preoperative thoracic CT angiography showing pulmonary embolization (PE) on the inferior branch of the left pulmonary artery (arrow). (D) MRI imaging that shows a central mass enhancement (arrow) on the Eustachian valve.

mass did not decrease. An MRI study confirmed the size and location of the mass in the RA and provided further evidence of central enhancement, which raised the suspicion of tumoral lesions (Figure 1D). After consulting with the local Heart Team, the decision to proceed with surgical treatment was made in order to prevent pulmonary embolization and obtain a definite diagnosis of this RA mass.

Under general anesthesia, we proceeded with femoral arterial and venous cannulation, followed by the initiation of cardiopulmonary bypass. We then performed a re-sternotomy and carefully released pericardial adhesions. A second venous cannula was inserted into the superior vena cava. The operation proceeded under normothermic cardiopulmonary bypass, and we operated on the beating heart through a right atriotomy.

The intraoperative findings revealed a 2.5 cm × 2.1 cm mass (Supplementary Video S1; Figures 2A,B) arising from the Eustachian valve and a preoperatively undiagnosed Chiari network in RA. Both structures were resected and sent for a frozen section procedure, which excluded a malignancy. The final histopathological analysis described fibrotic tissues compatible

with an organized thrombus. The patient was discharged on postoperative day 7 without any complications.

Discussion

The present case describes a right atrial thrombotic mass attached to a very unusual structure, the Eustachian valve, which was discovered during a routine follow-up 18 months after his initial heart surgery which was performed with aorto-bicaval cannulation. During the resection of the RA mass, an extensive and unreported Chiari network was found and removed simultaneously.

The Eustachian valve (EV) is localized around the orifice of the inferior vena cava and right atrial junction. It is a remnant of the intrauterine fetal circulation that helps in directing oxygenated blood from the placenta into the left atrium through the right atrium and patent foramen ovale (1). It usually regresses during childhood. A persistent EV is a frequent finding in patients with

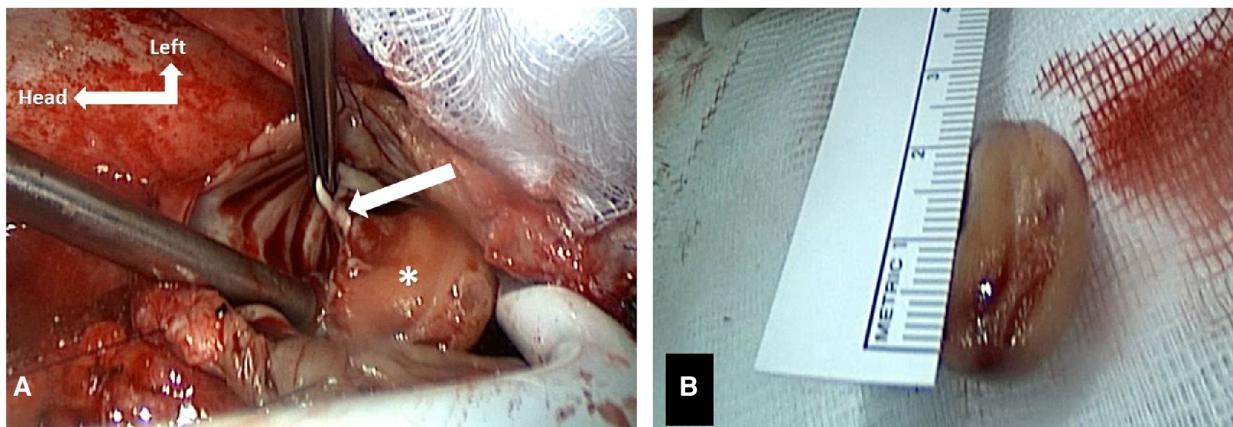


FIGURE 2
(A) Intraoperative findings of the right atrial mass attached to the Eustachian valve (arrow). (B) Intraoperative resected mass measuring approximately 2.5 cm x 2.1 cm.

a patent foramen ovale (2), as observed in the current reported case.

The Chiari network is occasionally seen in the RA near the opening of the inferior vena cava and the coronary sinus. First described by Hans Chiari in 1897, its prevalence has been variably reported to range between 2% and 13.6% (3–5). It may be associated with thrombi formation (6). It is a fenestrated, net-like structure and akin to the Eustachian valve, represents an embryonic remnant of the sinus venosus valve. The Chiari network is often incidentally diagnosed and may create turbulent flow and cause repetitive pulmonary thromboembolism.

In the presented case, the formation of the thrombus on the EV may be related to the presence of a Chiari network. Another possibility of the formation of the thrombus could be explained by endothelial trauma during the inferior vena cava cannulation in the first operation.

The combination of persistent EV and a Chiari network is a critical combination predisposing to thrombus formation in the RA. Embolization of parts of the RA thrombus is likely to be the source of the diagnosed pulmonary emboli in our patient. Additionally, deep vein thrombosis was ruled out through a dedicated Doppler exam.

Oral anticoagulant treatment is considered the first-line treatment in the presence of intra-cardiac thrombi (7). Because of the size of the RA, the mass did not decrease following 2 months of oral anticoagulant treatment, and suspicion of an intra-cardiac tumor was raised. This suspicion was further supported by central mass contrast uptake observed in the MRI study (Figure 1D). Cardiac MRI can be used to differentiate between intra-cardiac thrombi and tumors (8). Thrombi typically do not show contrast uptake and appear dark on LGE (late gadolinium enhancement), with a surrounding area of high uptake from adjoining blood or myocardium. Conversely, tumors exhibit central contrast uptake and delayed enhancement, as observed in our case, thus prompting surgical treatment. Although cardiac MRI has a high accuracy, in this particular case, it produced a false positive result (9).

Conclusion

A right atrial thrombotic mass attached to the Eustachian valve discovered during postoperative follow-up is a rare finding and may be misdiagnosed as a potential malignancy. Although imaging studies are useful for the initial and differential diagnosis of RA masses, obtaining a final diagnosis without surgery is not always possible. In case of a suspicion of a malignant process or a possible source of repetitive pulmonary embolism, surgical resection is necessary. Our case highlights the importance of continuous postoperative follow-up including regular imaging exams, even in asymptomatic patients. Swift diagnosis, treatment, and resection may help prevent undesirable outcomes.

Author's note

Parts of the content of this manuscript were presented in June 2021 at the Joint Annual Meeting of the Swiss Society of Cardiology (SSC) and the Swiss Society of Cardiac and Thoracic Vascular Surgery (SSCC), and part of its content was included in the Conference Abstract Booklet.

Data availability statement

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

Ethics statement

Ethical approval was not required for the studies involving humans because in accordance with our local Ethics Committee,

case reports with <5 patients are waived of ethical approval. Moreover, the information provided is completely de-identified. 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

JJ: Conceptualization, Data curation, Formal analysis, Investigation, Writing – original draft. JM: Formal analysis, Writing – review & editing, Investigation, Methodology. HM: Supervision, Validation, Writing – review & editing. MC: Formal analysis, Writing – review & editing, Supervision, Validation. CH: Formal analysis, Supervision, Validation, Writing – review & editing, Conceptualization, Visualization.

Funding

The author(s) declare financial support was received for the research, authorship, and/or publication of this article.

Open access funding by University of Geneva.

References

1. Gray's Textbook of Anatomy. 38th ed. 1995, pg. 1477.
2. Schuchlenz HW, Saurer G, Weihs W, Rehak P. Persisting eustachian valve in adults: relation to patent foramen ovale and cerebrovascular events. *J Am Soc Echocardiogr.* (2004) 17(3):231–3. doi: 10.1016/j.echo.2003.12.003
3. Chiari H. Ueber Netzbildungen im rechten Vorhofedes Herzens. *Beitr Pathol Anat.* (1897) 22:1–10.
4. Bhatnagar KP, Nettleton GS, Campbell FR, Wagner CE, Kuwabara N, Muresian H. Chiari anomalies in the human right atrium. *Clin Anat.* (2006) 19:510–6. doi: 10.1002/ca.20195
5. Yater WM. The paradox of Chiari's network. Review and report of a case of Chiari's network ensnaring a large embolus. *Am Heart J.* (1936) 11:542–52. doi: 10.1016/S0002-8703(36)90472-6
6. Schneider B, Hofmann T, Justen MH, Meinertz T. Chiari's network: normal anatomic variant or risk factor for arterial embolic events? *J Am Coll Cardiol.* (1995) 26:203–10. doi: 10.1016/0735-1097(95)00144-O
7. Islam M, Nesheim D, Acquah S, Kory P, Kouroumi I, Ramesh N, et al. Right heart thrombi: patient outcomes by treatment modality and predictors of mortality: a pooled analysis. *J Intensive Care Med.* (2019) 34(11–12):930–7. doi: 10.1177/0885066618808193
8. Pazos-López P, Pozo E, Siqueira ME, García-Lunar I, Cham M, Jacobi A, et al. Value of CMR for the differential diagnosis of cardiac masses. *JACC Cardiovasc Imaging.* (2014) 7(9):896–905. doi: 10.1016/j.jcmg.2014.05.009
9. Weinsaft JW, Kim RJ, Ross M, Krauser D, Manoushagian S, LaBounty TM, et al. Contrast-enhanced anatomic imaging as compared to contrast-enhanced tissue characterization for detection of left ventricular thrombus. *JACC Cardiovasc Imaging.* (2009) 2(8):969–79. doi: 10.1016/j.jcmg.2009.03.017

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.

Publisher's note

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

Supplementary material

The Supplementary Material for this article can be found online at: <https://www.frontiersin.org/articles/10.3389/fcvm.2023.1268918/full#supplementary-material>

SUPPLEMENTARY VIDEO S1

Video of the surgical procedure, showing the right atrial mass attached to the Eustachian valve and its removal.



OPEN ACCESS

EDITED BY

Giuseppe Gatti,
Azienda Sanitaria Universitaria Giuliano Isontina,
Italy

REVIEWED BY

Palleti Rajashekhar,
All India Institute of Medical Sciences, India
Luca Barozzi,
Integrated University Hospital Verona, Italy

*CORRESPONDENCE

Qi Miao
✉ miaoqipumc@hotmail.com

¹These authors have contributed equally to this work

RECEIVED 12 June 2023

ACCEPTED 25 October 2023

PUBLISHED 28 November 2023

CITATION

Chu P, Tang Y, Liu X and Miao Q (2023) Case Report: Pericardial patch repair of mitral annulus and mitral valve for a left atrial dissection caused by parasitic infective endocarditis.

Front. Cardiovasc. Med. 10:1239019.
doi: 10.3389/fcvm.2023.1239019

COPYRIGHT

© 2023 Chu, Tang, Liu and Miao. This is an open-access article distributed under the terms of the [Creative Commons Attribution License \(CC BY\)](#). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

Case Report: Pericardial patch repair of mitral annulus and mitral valve for a left atrial dissection caused by parasitic infective endocarditis

PeiShan Chu^{1,2†}, Yi Tang^{1,2†}, XinPei Liu^{1,2} and Qi Miao^{1*}

¹Department of Cardiac Surgery, Peking Union Medical College Hospital, Dongcheng District, Beijing, China, ²Chinese Academy of Medical Sciences & Peking Union Medical College, Beijing, China

Introduction: Left atrial dissection is a rare event, typically associated with cardiac manipulation. We report the first case of a left atrial dissection caused by parasitic infectious endocarditis, which required the use of patch repair for the damaged mitral annulus and valve.

Case Presentation: To treat heart failure in a 43-year-old man with left atrial dissection, we performed a patch repair of the mitral annulus and valve using autologous pericardium.

Conclusion: We encourage novel surgery for complicated infectious endocarditis.

KEYWORDS

left atrial dissection, parasite, infectious endocarditis, patch repair of mitral annulus and valve, patch repair for left atrial dissection

Introduction

Left atrial dissection (LatD) is an extremely rare condition characterized by a false blood-filled cavity or lumen extending from the mitral annulus (MA) to the left atrium (LA) (1, 2). It is often linked to cardiac manipulation, myocardial infarction (MI) and blunt cardiac trauma (1, 2). Spontaneous LatD are associated with conditions such as cardiac amyloidosis, severe mitral annular calcification, and infectious endocarditis (IE) in some studies (1, 2). The majority of IE cases are caused by bacterial or fungal infestation (1–3). Here, we present a unique LatD case caused by parasitic infestation of heart, along with the first report using pericardial patch repair for posterior mitral annulus in a LatD associated with IE (1–4). To prevent reinfection, we utilized autologous pericardial patch to repair MA and mitral valve (MV).

Case presentation

A 43-year-old Chinese man with no prior history of cardiac surgery or trauma complained diarrhea, general fatigue, and lower extremities edema for three months. Laboratory test revealed an absolute eosinophil count of $5.69 \text{ cells} \times 10^9/\text{L}$ with 37.8%

Abbreviations

LatD, left atrial dissection; LA, left atrial; MI, myocardial infarction; IE, infectious endocarditis; MA, mitral annulus; MV, mitral valve; LV, left ventricle; PML, posterior mitral leaflet.

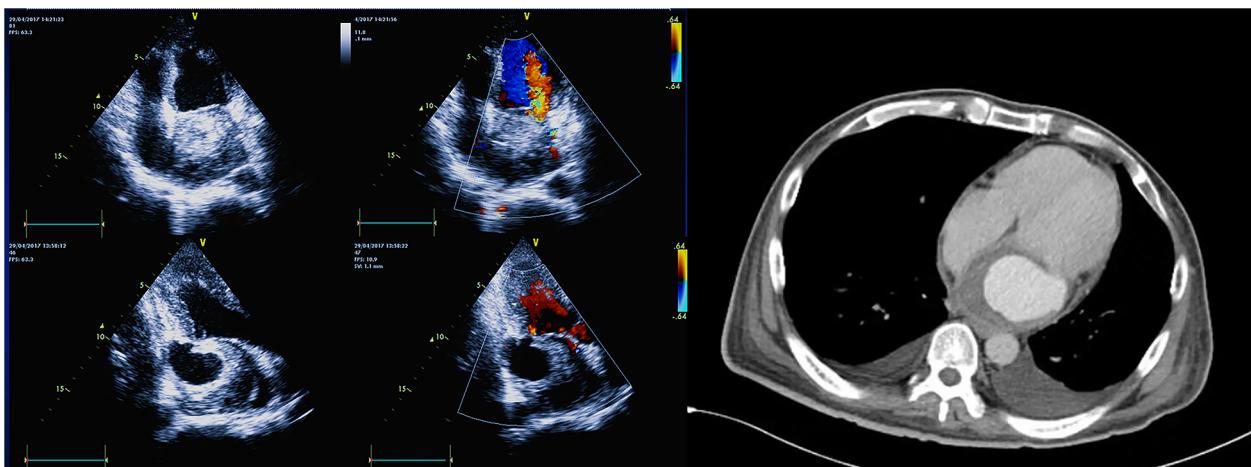


FIGURE 1

TTE showed a mass containing thrombus in the LA and a suspicious endocardial orifice beneath the root of the PML on the LV. CT revealed contrast filling the mass cavity.

eosinophils. Transthoracic echocardiogram (TTE) showed a 5.4 × 6.0 cm mass containing thrombus in the posterior wall of the left atrium (LA), causing functional mitral insufficiency and MV obstruction. In addition, there was a suspicious endocardial orifice beneath the root of the posterior mitral leaflet (PML) on the left ventricle (LV). Enhanced Computed tomography (CT) revealed the contrast filling the mass cavity, indicating intramural hematoma. (Figure 1) Left atrial dissection caused by parasitic infestation was suspected. Based on the patient's hemodynamic collapse, emergency surgery was necessary.

The surgical procedures were recorded in **Supplementary Material Video 1**. By removing the thrombus and debris from the false cavity and excising the dissected endocardium, the surgeon found an orifice on the left ventricle beneath the root of

the PML, which was confirmed as the entry of the LatD. Direct incision was performed in the P2, and the posterior MA was cut through to reach the orifice. An autologous pericardial patch with an ellipse shape was employed to reconstruct the defect of the LV and MA. After reaching the proposed neo-annulus line, the rest of the ellipse was used to augment the PML. An annuloplasty was performed with another pericardial band (refer to Figure 2). The reconstructed MV performed well under TEE after weaning from the cardiopulmonary bypass.

Although the specimen produced no findings, the eosinophil count and the percentage of eosinophils were sharply decreased after surgery. The stool examination revealed the ova of *Clonorchis sinensis*, *Metagonimus yokogawai* and *Echinostoma* (refer to Figure 3), confirming a LatD associated with parasitic infestation of heart. After completing anti-parasitic treatment, the patient was discharged and remained healthy during the 2-year follow-up period (5) (refer to Table 1).

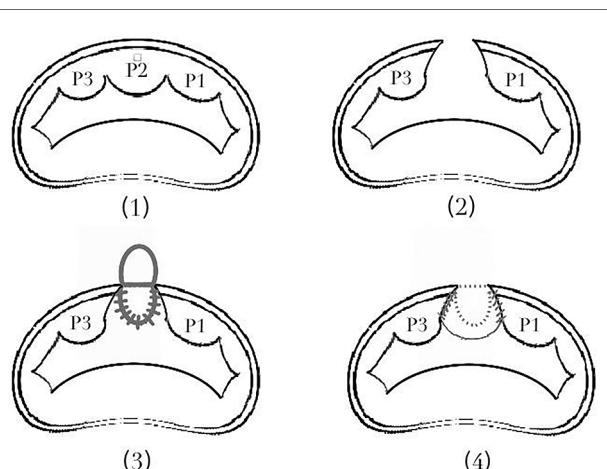


FIGURE 2

The procedure of patch repair for defect of LV, MV and MA. (2) To reach the orifice, we direct incised the P2, and cut through the posterior MA. (3) An ellipse-shape autologous pericardial patch was employed to reconstruct the defect of the LV and MA. (4) After reaching the proposed neo-annulus line, the rest of the ellipse was used to augment the PML.

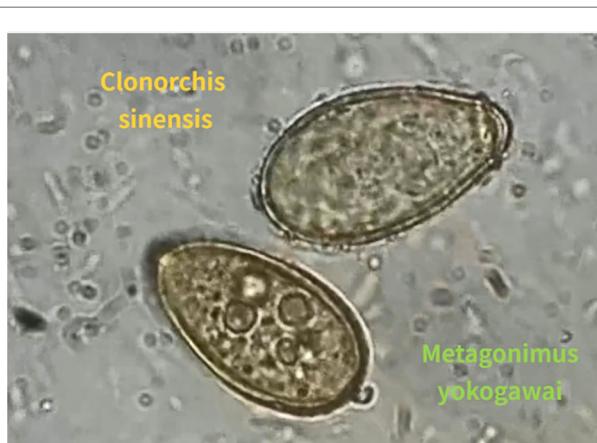


FIGURE 3

The ova of *clonorchis sinensis* (orange) and *metagonimus yokogawai* (green) in the stool examination.

TABLE 1 Timeline.

Time	Situation
3 Month Previously	Onset of diarrhea, general fatigue, and lower extremities edema
Initial Presentation	TTE showed a 5.4×6.0 cm mass in the posterior wall of the LA, causing functional mitral insufficiency and MV obstruction, and a suspicious endocardial orifice in the LV. Diagnosis of left atrial dissection was suspicious made. Hemodynamic instability required emergency surgery.
Surgery Day	Pericardial patch repair of mitral annulus and mitral valve was performed
2 weeks later	A stool examination revealed the oval of parasites. Anti-parasitic treatment was performed.
5 months later	Anti-parasitic treatment was finished. A stool examination showed negative.
2 years later	Patient remained healthy.

Discussion

Left atrial dissection can result from various cardiac interventions, including both surgical procedures (primarily MV surgery) and catheter-based interventions (1, 2). Cases of LatD resulting from MI and cardiac trauma have also been recorded (1, 2). Fukuhara et al. reported only 7 cases of spontaneous LatD, one of which was related to IE without cardiac manipulation (1). With another case of LatD with IE in other report, both patients were caused by bacterial infection and died before emergency surgery could be performed (1–3). This might be the first report of LatD resulting from parasitic infestation of heart and survived after surgery.

Transesophageal echocardiography (TEE) is the preferred method for diagnosing LatD, but its efficacy and diagnostic value are still limited (1). Echocardiogram often shows a left atrial mass or a tamponade, particularly when there is thrombus attached (1). Visualizing the entry of the dissection or communication between the inflow and the LA can be challenging (1). A TTE was performed in the emergency room on our patient and revealed an ambiguous left atrial mass with regurgitation flow under the PML, which could have been misdiagnosed as a leaflet perforation. Enhanced CT imaging is also limited in identifying precisely when the thrombus filled in the false cavity.

Hemodynamic instability is mainly secondary to obstruction of MV inflow or pulmonary vein orifice and eventually becomes congestive heart failure and low-output syndrome, making conservative management impractical. Surgery was necessary for our patient (2).

To achieve successful repair, three main keys are the following: (a.) to gain adequate evacuation of the hematoma; (b.) obliteration of the false lumen; and (c.) addressing the entry point, if identified. If the communication is not repaired, the dissection may recur under persistent pressurized inflow (1, 2). Therefore, we debride thrombus and resect the dissected endocardium. To close the orifice in the LV, a posterior annulus cutting was necessary. However, the tissue of LA and MA is often delicate and should be preserved whenever possible.

Encouraged by Masashi's successful application of the technique for patch repair of mitral annulus calcification, we proceeded with posterior MA cutting and patch repair (6). We chose the autologous pericardium as patch to prevent artificial graft reinfection.

Conclusion

By introducing this rare parasitic infestation of LatD and its surgical details, we aim to encourage novel surgery for complicated infective endocarditis.

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

This study was approved by the Institutional Review Board (IRB). Informed written consent was obtained from the patient. 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

PC drafted the manuscript and drew the figure. YT clipped the video. XL revised the manuscript. QM supervised the report. All authors contributed to the article and approved the submitted version.

Funding

This work was supported by the National High Level Hospital Clinical Research Funding Grant No. 2022-PUMCH-B-104.

Acknowledgments

I would like to thank QM and XL who taught me every aspect of mitral valve and annulus repair as well as mitral annuloplasty, and YT for the video editing.

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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated

organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

Supplementary material

The Supplementary Material for this article can be found online at: <https://www.frontiersin.org/articles/10.3389/fcvm.2023.1239019/full#supplementary-material>

References

1. Fukuhara S, Dimitrova KR, Geller CM, Hoffman DM, Tranbaugh RF. Left atrial dissection: an almost unknown entity. *Interact Cardiovasc Thorac Surg.* (2015) 20 (1):96–100. doi: 10.1093/icvts/ivu317
2. Gallego P, Oliver JM, Gonzalez A, Dominguez FJ, Sanchez-Recalde A, Mesa JM. Left atrial dissection: pathogenesis, clinical course, and transesophageal echocardiographic recognition. *J Am Soc Echocardiogr.* (2001) 14(8):813–20. doi: 10.1067/mje.2001.113366
3. Saad M, Isbitan A, Roushdy A, Shamoony F. Left atrial wall dissection: a rare sequela of native-valve endocarditis. *Tex Heart Inst J.* (2015) 42(2):178–80. doi: 10.14503/THIJ-13-3989
4. Cordero Lorenzana ML, López Pérez JM, Merayo Macías E, Gulías López JM, Paz Rodríguez J. Disección auricular izquierda y endocarditis infecciosa. *Rev Esp Cardiol.* (1998) 51:402–3. doi: 10.1016/S0300-8932(98)74765-7
5. Li L, Liu X, Zhou B, Zhang S, Wang G, Ma G, et al. Multiple food-borne trematodiases with profound systemic involvement: a case report and literature review. *BMC Infect Dis.* (2019) 19(1):526. doi: 10.1186/s12879-019-4140-y
6. Ura M. Mitral valve repair with decalcification of the annulus and pericardial patch repair via the trans-septal approach. *Ann Cardiothorac Surg.* (2015) 4 (5):474–5. doi: 10.3978/j.issn.2225-319X.2015.04.04



OPEN ACCESS

EDITED BY

Giuseppe Gatti,
Azienda Sanitaria Universitaria Giuliano
Isontina, Italy

REVIEWED BY

George Samanidis,
Onassis Cardiac Surgery Center, Greece
Omar R. J. Tamimi,
King Fahd Medical City, Saudi Arabia

*CORRESPONDENCE

Xuejun Li
✉ gybj2005@qq.com
Siyuan Yang
✉ 15772955@qq.com

¹These authors share first authorship

RECEIVED 02 November 2023

ACCEPTED 07 December 2023

PUBLISHED 20 December 2023

CITATION

Wu K, Fan X, Hu X, Li X and Yang S (2023) Case Report: Surgical management of idiopathic pulmonary aneurysms and review surgical approaches. *Front. Cardiovasc. Med.* 10:1331982. doi: 10.3389/fcvm.2023.1331982

COPYRIGHT

© 2023 Wu, Fan, Hu, Li and Yang. This is an open-access article distributed under the terms of the [Creative Commons Attribution License \(CC BY\)](#). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

Case Report: Surgical management of idiopathic pulmonary aneurysms and review surgical approaches

Kui Wu^{1†}, Xuan Fan^{2†}, Xuanyi Hu¹, Xuejun Li^{1*} and Siyuan Yang^{1*}

¹Department of Cardiovascular Surgery, The Affiliated Hospital of Guizhou Medical University, Guiyang, China, ²Department of Surgery, Guizhou Provincial Corps Hospital of Chinese People's Armed Police Forces, Guiyang, China

Idiopathic pulmonary aneurysm is a clinically rare condition characterized by an unknown etiology and episodic occurrence. Despite its rarity, idiopathic pulmonary artery aneurysm poses potential risks to patients. Currently, there is a lack of established clinical guidelines and consensus regarding its management, leading to ongoing controversies in treatment strategies. Particularly, the optimal approach for addressing the main pulmonary artery, its branches, and the pulmonary artery valve remains uncertain. A 57-year-old female patient presented with chest pain and tightness, leading to the diagnosis of idiopathic pulmonary artery aneurysm after excluding other potential causes. Subsequently, she underwent surgical treatment. However, during the surgery, the pulmonary artery wall was found to be extremely weak, prompting us to employ a surgical approach involving the utilization of autologous vessel wrapping with artificial grafts. By summarizing almost all surgical treatment strategies reported in recent years, including the management of pulmonary artery vessels and the pulmonary valve, we have developed a treatment flow chart. This flowchart serves as a valuable guide for the management of future cases presenting similar challenges, offering clinicians valuable insights and evidence-based recommendations.

KEYWORDS

pulmonary aneurysms, idiopathic, procedure, vascular replacement, case report

1. Introduction

Pulmonary artery aneurysms (PAAs) are considered to be a rare disease. Deterling and Clagett (1) discovered 8 cases of PAAs in a series of 109,571 consecutive postmortem examinations, resulting in a prevalence rate of 0.0073%. Regarding the definition of the size of pulmonary aneurysms, a population-based survey by Berger et al. (2) reported that the mean diameter of pulmonary arteries (PAs) in a healthy population was 32.0 ± 4.6 mm and suggested that the threshold definition of PAAs should not be less than 45 mm. The etiology of PAAs can be diverse and encompass various factors. These include congenital heart defects, connective tissue abnormalities (e.g., Marfan's syndrome), infectious diseases (e.g., syphilis, tuberculosis, suppurative bacterial infections, and fungal pneumonia), vasculitis (e.g., leukoaraiosis), idiopathic pulmonary arterial hypertension, chronic pulmonary embolisms, neoplasms (e.g., primary lung cancers and lung metastases, medically induced cardiac surgery, as well as the unexplained (idiopathic) cause (3).

Idiopathic PAAs is a rare and mysterious condition characterized by an aneurysm in the pulmonary artery without any known cause. To offer a more precise characterization of idiopathic pulmonary aneurysms, four distinct pathological criteria have been established. (i) dilation of the pulmonary trunk, with or without dilation of other arteries; (ii) the absence of abnormal shunts within or outside the heart; (iii) the absence of chronic cardiorespiratory disease confirmed through clinical or autopsy findings; and (iv) the absence of arterial diseases like syphilis, significant atherosclerosis, or small arteriosclerosis (4). The current diagnostic criteria require the exclusion of these underlying causes to determine the presence of pulmonary artery dilation. Idiopathic PAAs typically manifest as asymptomatic, occasionally present with hemoptysis, and have been reported to result in compression of coronary vessels or the superior vena cava (5–7). For proximal pulmonary aneurysms, conservative treatment is usually used. As aneurysm size increases, surgical resection or graft repair may need to be considered.

Currently, there are no established guidelines for the diagnosis and treatment of PAAs. Although, some literature suggests that conservative treatment has shown favorable clinical outcomes (8, 9). However, large pulmonary aneurysms still pose significant risks, and surgical intervention has been found to provide considerable clinical benefits for patients. In this report, we present a case of surgical treatment for idiopathic PAAs, providing a detailed account of the treatment course and outcomes. Additionally, we review the existing literature on the surgical approach to proximal pulmonary aneurysms.

2. Case presentation

A 57-year-old woman visited community hospital due to experiencing chest tightness and chest pain for over a year. A chest computed tomography (CT) scan revealed aneurysmal dilation of the pulmonary artery. She had a history of tuberculosis over 20 years ago but stated that she had been successfully treated and cured. During the examination, the patient's blood pressure was measured at 110/70 mmHg, and her pulse rate was recorded at 73 beats per minute. She has a height of 155 cm and a weight of 45 kg. The computed tomography angiography (CTA) scan of the pulmonary arteries revealed significant aneurysmal dilation of the main pulmonary artery and the bifurcation lumen (about 5.6 cm at the wider part), which was considered to be a pulmonary aneurysm; the walls of the main pulmonary artery and its branches appeared smooth and continuous, with no apparent filling defect within the lumen (Figures 1A,B). The electrocardiogram (ECG) displayed a normal sinus rhythm. The echocardiogram reveals no abnormalities in the valves, and there are no abnormal *in vivo* or *in vitro* shunts present. Additionally, there is aneurysmal dilatation of the pulmonary arteries (Supplementary Figure S1). Right heart catheterization revealed a pulmonary artery pressure of 21/11/14 mmHg. Coronary angiography suggests no abnormality. Relevant preoperative routine blood tests are shown in Supplementary Table S1, which were mainly positive for Mycobacterium tuberculosis CD4+ T cells. We recommended

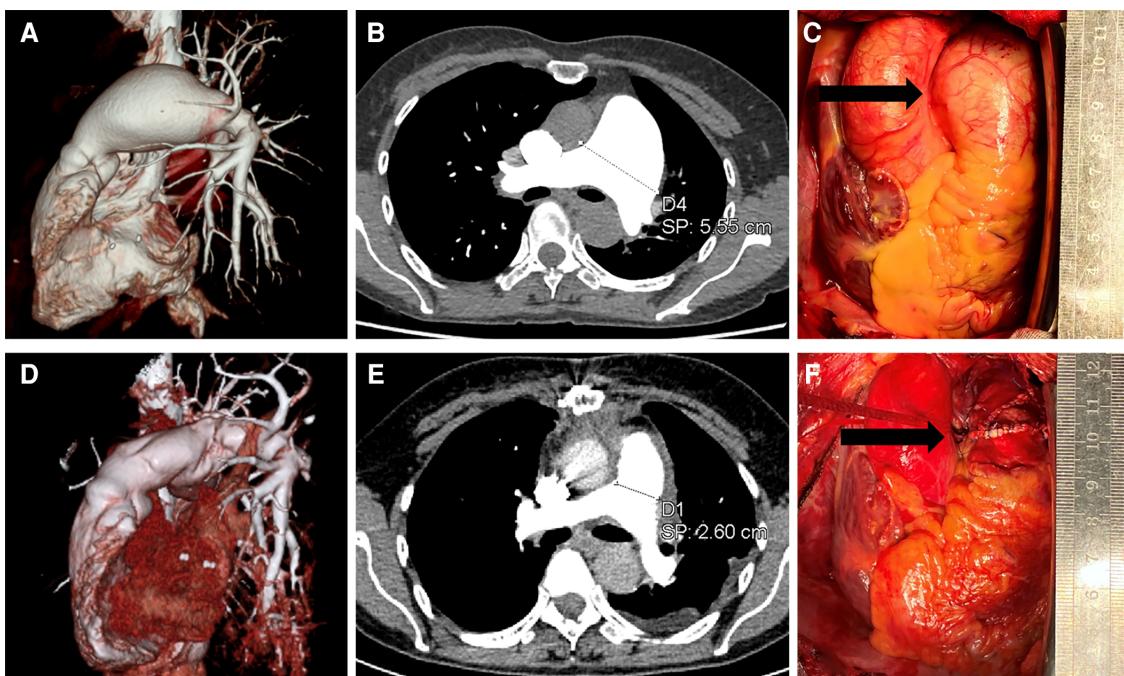


FIGURE 1

Preoperative and postoperative data. (A,B) are preoperative pulmonary artery CTA images showing an aneurysmal dilation of the pulmonary artery, with a maximum width of 5.55 cm. (D,E) are postoperative pulmonary artery CTA images demonstrating restoration of normal diameter in the main pulmonary artery, approximately 2.6 cm. Images (C,F) are intraoperative images, with (C) showing the pulmonary artery aneurysm under direct visualization, and (F) displaying the pulmonary artery after vessel replacement.

surgical treatment of the pulmonary aneurysm. Intraoperatively, it was noted that the main pulmonary artery exhibited significant dilatation from 2 cm above its origin to the bifurcation of the right and left pulmonary arteries, with a maximum diameter of approximately 5.5 cm, and no palpable thrill was detected at the root, and there was a slight dilation observed in the left and right pulmonary arteries (Figure 1C). Cardiopulmonary bypass was initiated, and a sequential blockade of the superior vena cava and inferior vena cava, as well as the right and left pulmonary arteries, was implemented. After the aortic occlusion, cardiac arrest is induced by the infusion of a specialized cardiac arrest solution, effectively ceasing the heart's activity. Intraoperative probes reveal that the pulmonary arteries have thin walls without any signs of dissection or thrombosis. The pulmonary valve annulus is not dilated, and there is no significant regurgitation observed. The aneurysm of the main pulmonary artery was dissected longitudinally and replaced using an artificial blood vessel (24 mm in diameter), which was anastomosed to the proximal and distal ends of the main pulmonary artery, respectively, and the patient's own dilated portion of the pulmonary artery wall was partially excised. The artificial blood vessel was re-sutured and wrapped (Figure 1F). Postoperatively, the patient was returned to the intensive care unit. The patient was transferred to a regular ward on the second day after the surgery and was discharged smoothly one week postoperatively. The diseased vessel was excised and sent for routine pathological examination. Pathological findings showed thinning of the pulmonary artery wall with no other significant abnormalities (Supplementary Figure S2). Postoperative follow-up pulmonary artery CTA showed that the diameter of the main pulmonary artery returned to normal (Figures 1D,E). Anticoagulation with warfarin for 6 months. The patient came for a follow-up visit at the outpatient clinic 2 months later, reporting satisfactory recovery and no specific complaints of discomfort.

3. Discussion

In our clinical practice, we have encountered patients with pulmonary aneurysms whose causes could be determined, including two cases of pulmonary aneurysms caused by congenital heart disease (Supplementary Figure S3). However, idiopathic pulmonary aneurysms are considered to be rare. The patient was admitted to the hospital due to chest pain, and subsequent examination eliminated the potential cause of a pulmonary aneurysm. The patient had a previous history of tuberculosis over two decades ago, which had resolved over time. Upon admission, the patient's CD4+ T-cell test for tuberculosis antigen specificity yielded positive results, indicating a prior tuberculosis infection. However, it is important to note that pulmonary aneurysms resulting from tuberculosis infection typically manifest in the distal pulmonary arteries and are not commonly associated with enlargement of the main pulmonary arteries (10). Throughout the surgical procedure, we observed the following details. (i) Preoperative evaluation of the thickness of the pulmonary artery aneurysm vessels is necessary. We observed

that the pulmonary arteries exhibited a marked thinness, measuring approximately 1 mm. Despite the absence of elevated pulmonary artery pressure, we chose to use the patient's blood vessel to wrap the artificial blood vessel as a precautionary measure for enhanced safety. Nonetheless, during the distal anastomosis, we noted that the vessel walls remained susceptible to tearing due to their thin and fragile condition. Consequently, we fortified the vessel walls by applying an external vascular patch. (ii) To mitigate the risk of bleeding, we advised complete exposure of the bilateral pulmonary arteries before the surgery and temporary occlusion using polyester tapes. This approach facilitated a more optimal surgical field of vision. (iii) Regarding whether to stop the heartbeat, we opted for a "stopped" approach during the surgery to ensure a clearer view for the patient, considering the blood reflux from the coronary sinus. (iv) Regarding the selection of anticoagulation therapy, our standard practice following ascending aorta artificial graft surgery involves the administration of aspirin for 3–6 months. However, considering the slower blood flow velocity in the pulmonary artery compared to the aortic side and the absence of comprehensive anticoagulation protocols in existing literature, we have opted for a warfarin anticoagulation regimen for 3–6 months to prioritize patient safety.

We conducted a comprehensive analysis of recent cases involving idiopathic pulmonary aneurysms and examined the surgical techniques employed in these cases (Supplementary Table S2). Previous studies primarily focused on discussing the indications for surgery, with limited discussion on the available surgical treatment options and choices. By integrating patient cases and conducting a thorough literature review, we have compiled a summary of the various surgical treatment options and choices available for this condition.

The initial step is to assess whether the patient meets the necessary indications for surgery. For indications of surgery for pulmonary aneurysms, Kreibich et al. (3) suggest the following factors be considered: (I) absolute aneurysm diameter ≥ 5.5 cm; (II) increase in aneurysm diameter of ≥ 0.5 cm within 6 months; (III) compression of adjacent structures; (IV) formation of a thrombus within the aneurysm; (V) development of clinical symptoms; (VI) presence of valvulopathy or shunts; (VII) confirmed diagnosis of pulmonary arterial hypertension; and (VIII) rupture of aneurysm or sandwich formation. The second step involves managing the pulmonary artery vasculature. Firstly, the thickness of the vessel wall needs to be assessed. For patients with appropriate or normal vessel wall thickness, direct removal of the pulmonary aneurysm can be performed. However, for patients with very thin pulmonary arteries, it is recommended to use artificial vascular grafts, along with wrapping using the patient's blood vessels (11–13). Subsequently, the involvement of both the right and left pulmonary arteries must be taken into consideration. In cases where the pulmonary arteries are cumulatively involved or bilaterally involved, simultaneous vascular replacement on one or both sides may be necessary (14–17) (Figure 2). The last step involves considering the management of the pulmonary valve. The functionality of the pulmonary valve needs to be evaluated to determine if it is

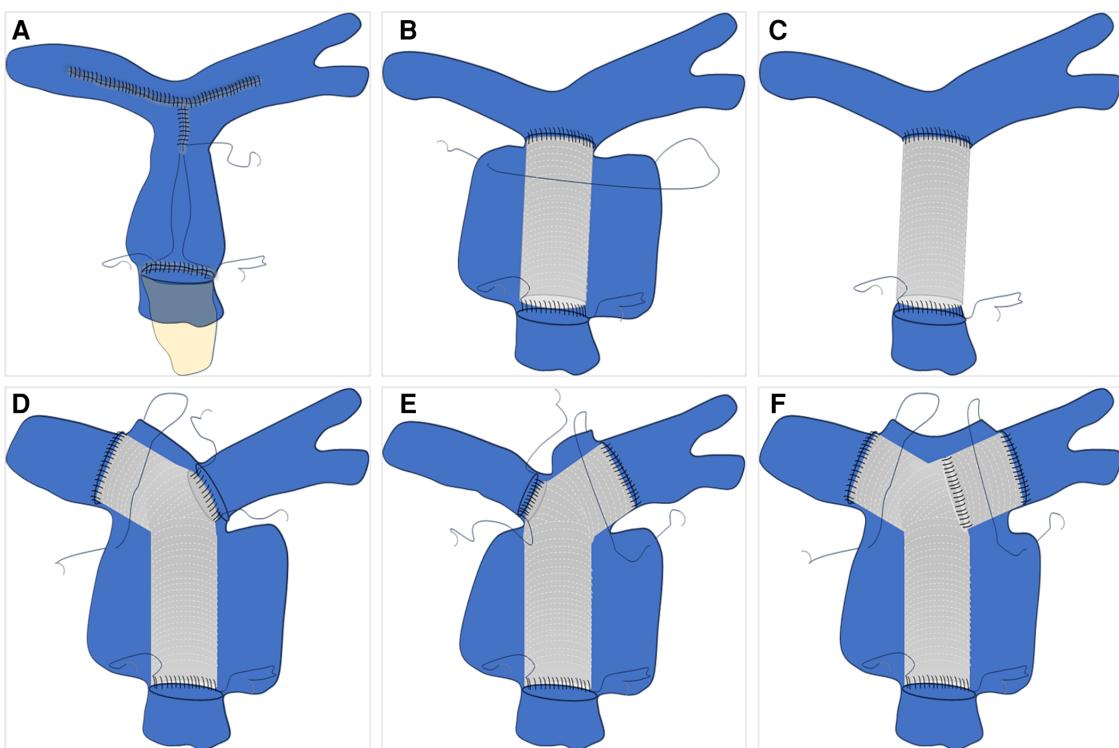


FIGURE 2

Pulmonary artery vascular management schematic. (A) Direct excision and suturing of the dilated pulmonary artery. (B) Replacement of the ascending pulmonary artery with an artificial blood vessel and wrapping. (C) Replacement of the ascending pulmonary artery with an artificial blood vessel without autologous vessel wrapping. (D–F) represent schematic illustrations of involvement of the left pulmonary artery, right pulmonary artery, or bilateral pulmonary artery branches, respectively, with subsequent replacement.

affected. If the valve function is normal and only the annulus is enlarged, a procedure similar to David's (16) or Devega's (15) can be performed, preserving as much function of the pulmonary valve as possible; if the valve function is abnormal, a bioprosthetic pulmonary valve is also a good option (12, 18) (Figure 3).

We have devised a treatment flowchart (Figure 4) exclusively applicable to patients diagnosed with idiopathic pulmonary aneurysm, excluding those with secondary pulmonary artery dilatation. This flowchart is intended to serve as a comprehensive guide for subsequent clinical management and decision-making processes.

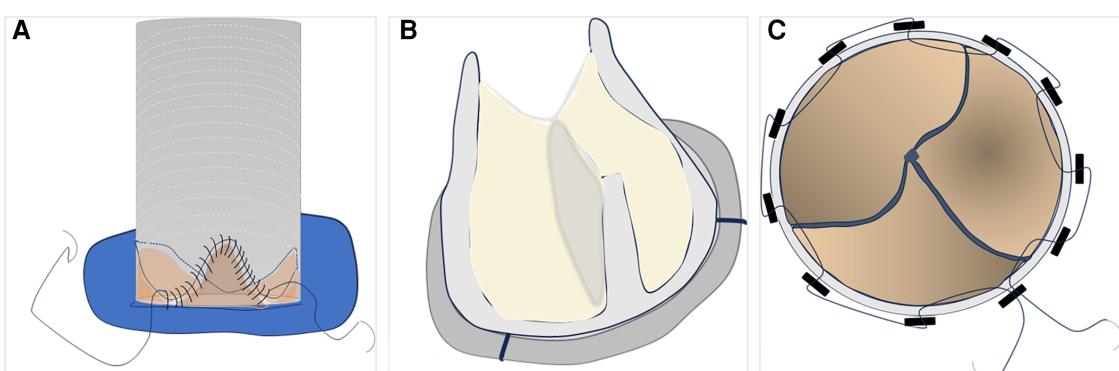


FIGURE 3

Schematic illustrations of pulmonary artery management. (A) Similar to the David procedure; (B) bioprosthetic valve replacement; (C) De Vega-like procedure.

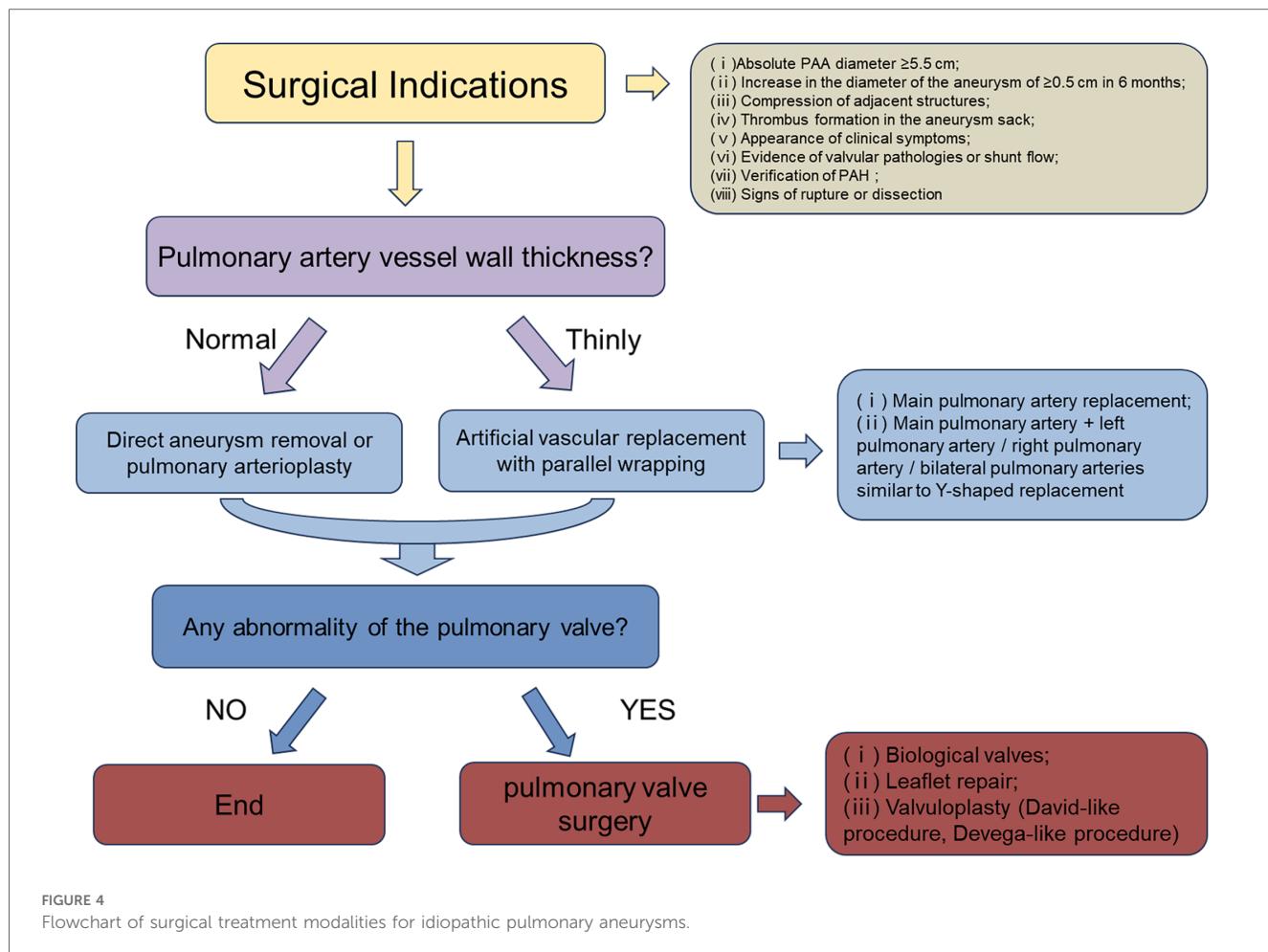


FIGURE 4
Flowchart of surgical treatment modalities for idiopathic pulmonary aneurysms.

4. Conclusion

Surgical treatment of Idiopathic PAAs is comparatively safe and clinically beneficial for postoperative patients. However, it requires attention to a series of details and selection of appropriate surgical techniques. By creating a flowchart, we can provide valuable clinical thinking and procedures for subsequent similar patients, thereby improving the accuracy and effectiveness of treatment and providing better medical care.

5. Patient perspective

"I had no prior knowledge about idiopathic pulmonary artery, the type of disease I was diagnosed with. However, I consider myself fortunate to have received timely surgical intervention, which ensured my safety. I am deeply grateful to the medical staff for their exceptional treatment. I sincerely hope that my experience can serve as a valuable reference for similar cases". We appreciate the patient's encouragement and support in reporting the article!"

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

Ethical approval was not required for the studies involving humans because The Affiliated Hospital of Guizhou Medical University IRB deems case reports of less than three patients not to constitute human subject research and therefore not to require IRB review and approval. Written informed consent was obtained from the families of the patient for scientific activity including publication of this case report. 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

KW: Writing – original draft. XF: Data curation, Software, Writing – original draft. XH: Writing – review & editing. SY: Writing – review & editing. XL: 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.

References

1. Deterling RA Jr, Clagett OT. Aneurysm of the pulmonary artery; review of the literature and report of a case. *Am Heart J.* (1947) 34:471–99. doi: 10.1016/0002-8703(47)90527-9
2. Berger T, Siepe M, Simon B, Beyersdorf F, Chen Z, Kondov S, et al. Pulmonary artery diameter: means and normal limits-assessment by computed tomography angiography. *Interact Cardiovasc Thorac Surg.* (2022) 34:637–44. doi: 10.1093/icvts/ivab308
3. Kreibich M, Siepe M, Kroll J, Hohn R, Grohmann J, Beyersdorf F. Aneurysms of the pulmonary artery. *Circulation.* (2015) 131:310–6. doi: 10.1161/CIRCULATIONAHA.114.012907
4. Greene DG, Baldwin ED, Baldwin JS, Himmelstein A, Roh CE, Cournand A. Pure congenital pulmonary stenosis and idiopathic congenital dilatation of the pulmonary artery. *Am J Med.* (1949) 6:24–40. doi: 10.1016/0002-9343(49)90004-2
5. Rupprecht H, Ghidau M, Ditterich D. Ruptured pulmonary artery aneurysm mimicking pulmonary embolism. *Thorac Cardiovasc Surg.* (2012) 60:491–2. doi: 10.1055/s-0031-1280070
6. Zhang M, Li Q, Wu QC, Jiang YJ. A large, idiopathic, right pulmonary artery aneurysm with superior vena cava compression. *Eur J Cardiothorac Surg.* (2011) 39:1077. doi: 10.1016/j.ejcts.2011.01.050
7. Li CH, Barros AJ, Carreras F, Subirana MT, Pons-Lladó G. Idiopathic pulmonary artery aneurysm compressing the left main coronary artery. *Eur Heart J Cardiovasc Imaging.* (2012) 13:696. doi: 10.1093/ehjci/jes045
8. Sa-Kong H, Seol SH, No TH, Park DH, Jeong NR, Jeong SJ, et al. Huge idiopathic pulmonary artery aneurysm. *Radiol Case Rep.* (2017) 12:236–9. doi: 10.1093/ehjci/jes045
9. Xie J, Qin Y, Liu J, Liang W. A rare giant idiopathic pulmonary artery aneurysm and its management: a case report. *Asian J Surg.* (2023) 46:4416–7. doi: 10.1016/j.asjsur.2023.04.088
10. Marak JR, Kumar T, Gara H, Dwivedi S, Rasmussen aneurysm: case series of a rare complication of pulmonary Tuberculosis. *Respir Med Case Rep.* (2023) 45:101897. doi: 10.1016/j.rmc.2023.101897
11. Flaifel M, Suresh Daniel R, Nakanishi H, Than CA, Shiakos G, Tzanavaros I. A novel approach for the treatment of pulmonary artery aneurysm repair using inclusion technique: a case report. *Cureus.* (2023) 15:e36456. doi: 10.7759/cureus.36456
12. Stevens M, Swan KW, Bommareddi S, Ali SO. Two giants: giant cell arteritis causing a giant pulmonary artery aneurysm. *JTCVS Tech.* (2023) 20:79–82. doi: 10.1016/j.xjtc.2023.04.016
13. Badders J, Roughneen P, Mohan N, Roughneen E. Pulmonary artery aneurysm: a rarity and surgical enigma. *Cureus.* (2023) 15:e38157. doi: 10.7759/cureus.38157
14. Qian Q, Subbian SK, Kofidis T. Total pulmonary artery replacement with an avals-gelweave conduit in a patient with giant pulmonary artery aneurysm with pulmonary regurgitation. *J Card Surg.* (2020) 35:1122–4. doi: 10.1111/jocs.14517
15. Hong Son PD, Tu VN, Uoc NH, Vo HL. Successful aneurysmorrhaphy for a giant idiopathic pulmonary artery aneurysm. *Innovations (Phila).* (2020) 15:275–8. doi: 10.1177/1556984520911667
16. Cirić R, Boysan E, Behlul Altunkeser B, Aygul N, Cagli K, Cagli K, et al. David's procedure for pulmonary artery aneurysm. *J Card Surg.* (2020) 35:942–5. doi: 10.1111/jocs.14480
17. Haj-Yahia S, Sbairi M, Bali K, Darwazah A, Othman W, Zaghari M, et al. Case report and management approach in idiopathic pulmonary arteries aneurysm. *J Cardiothorac Surg.* (2018) 13:110. doi: 10.1186/s13019-018-0791-9
18. Worku BM, de Angelis P, Wingo ME, Leonard JR, Khan FM, Hameed I, et al. Pulmonary artery aneurysms: preoperative, intraoperative, and postoperative findings. *J Card Surg.* (2019) 34:570–6. doi: 10.1111/jocs.14070

Publisher's note

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

Supplementary material

The Supplementary Material for this article can be found online at: <https://www.frontiersin.org/articles/10.3389/fcvm.2023.1331982/full#supplementary-material>



OPEN ACCESS

EDITED BY

Giuseppe Gatti,
Azienda Sanitaria Universitaria Giuliano
Isontina, Italy

REVIEWED BY

Francesco Patanè,
Azienda Ospedaliera Ospedali Riuniti Papardo
Piemonte, Italy
Massimo Baudo,
Lankenau Institute for Medical Research,
United States

*CORRESPONDENCE

Chuanli Ren
✉ renchl@163.com

¹These authors share first authorship

RECEIVED 10 October 2023

ACCEPTED 28 December 2023

PUBLISHED 12 January 2024

CITATION

Tang C, Gao X, Chen T, Shao J, Zhu T, Zheng X and Ren C (2024) Case Report: Acute cerebral infarction caused by left atrial and right ventricular myxoma received emergency operation. *Front. Cardiovasc. Med.* 10:1316063. doi: 10.3389/fcvm.2023.1316063

COPYRIGHT

© 2024 Tang, Gao, Chen, Shao, Zhu, Zheng and Ren. This is an open-access article distributed under the terms of the [Creative Commons Attribution License \(CC BY\)](#). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

Case Report: Acute cerebral infarction caused by left atrial and right ventricular myxoma received emergency operation

Chengbin Tang^{1,2†}, Xianglong Gao^{1,2†}, Tao Chen^{1,2}, Jun Shao^{1,2},
Tao Zhu^{1,2}, Xucai Zheng³ and Chuanli Ren^{4*}

¹Department of Cardiovascular Surgery, Northern Jiangsu People's Hospital Affiliated to Yangzhou University, Yangzhou, China, ²The Yangzhou Clinical Medical College of Xuzhou Medical University, Xuzhou, China, ³Department of Breast and Thyroid Surgery, The First Affiliated Hospital of University of Science and Technology of China, Hefei, China, ⁴Department of Laboratory Medicine, Clinical College of Yangzhou University, Yangzhou, Jiangsu, China

Cardiac myxoma is a rare etiology of ischemic stroke, especially in young people. We report a case of multiple myxomas in left atrium and right ventricle inducing acute cerebral infarction. No significant abnormalities were detected in the patient's preoperative laboratory examination. Following emergency surgery, the patient's prognosis was satisfactory, providing valuable empirical insight for the surgical management of acute cerebral infarction in individuals diagnosed with cardiac myxoma. Our objective is to heighten awareness regarding the evaluation and treatment of patients with acute cerebral infarction subsequent to early diagnosis of cardiac myxoma.

KEYWORDS

myxomas, neurologic symptoms, emergency operation, case report, cardiac tumors

Introduction

Cardiac myxomas account for between 50% and 85% of all cardiac tumors (1). Sixty percent to 80% of cardiac myxomas occur in the left atrium, usually attached to fossa ovalis, the right atrium and ventricle and valves are relatively rare (1, 2). Controversy surrounds the treatment of neurologic symptoms caused by cardiac myxoma, focusing on whether to prioritize removing cardiac tumors or addressing cerebral infarction first.

A 25-year-old woman came to our emergency center with repeated chest tightness, palpitations, asthma, occasional hand and foot numbness, and worsening symptoms during daily activities over a 2-month period. Dizziness, headache, nausea and vomiting occurred 1 day before hospital admission. A magnetic resonance imaging (MRI) of the head revealed multiple specks and flaky signals in the left cerebral hemisphere, suggesting the possibility of multiple acute cerebral infarctions (Figure 1, arrow). Intraoperative transesophageal ultrasound demonstrated a soft mass shadow attached to the anterior lobe of the mitral valve, which collided with the left ventricle during diastole. Additionally, another mass shadow was observed at the apex of the right ventricle with minimal activity (Figure 2, arrow below). Subsequently, a dark red jelly-like mass, sized approximately 5 × 3 cm in the left atrium and 3 × 2 cm in the right ventricle, was identified and completely excised (Figure 3). The histopathological examination of a biopsy specimen confirmed the presence of cardiac myxomas (Figure 4).

Ten hours after admission, the patient underwent an emergency operation in which a median sternal incision was made to gain access to the chest. Heparinization of the

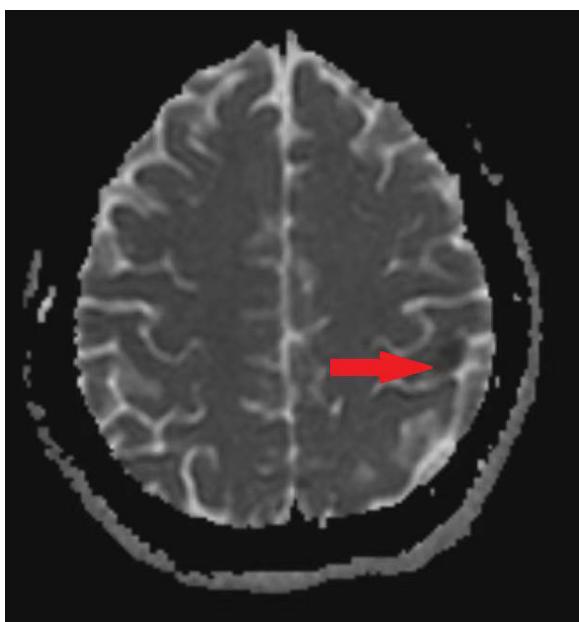


FIGURE 1
The MRI of the patient's head before operation, the arrow is the focus area.



FIGURE 3
The red jelly-like substance in the bowl is the tissue of myxoma removed by surgery.

entire body was then carried out, followed by the establishment of cardiopulmonary bypass through the ascending aorta and superior and inferior vena cava. Subsequently, the entire body was cooled, and the superior and inferior vena cava were blocked. The left atrium was then incised and drained, with temporary omission of the left atrial tube insertion, to prevent the myxoma from dislodging. The ascending aorta was then blocked and cardioplegia was infused through the aortic root before a longitudinal incision was made in the right atrium and atrial septum following cardiac arrest. During the procedure, a dark red jelly-like mass, measuring approximately 5×3 cm, was identified in the left atrium and successfully removed. A left

atrial retractor was then used to open the anterior lobe of the tricuspid valve, revealing a dark red jelly-like tumor measuring about 3×2 cm in the right ventricle. Following its removal, the tumor was meticulously rinsed multiple times with warm water to ensure no residual tumor was left in any cardiac cavity, thereby minimizing the risk of recurrence (3). The procedure

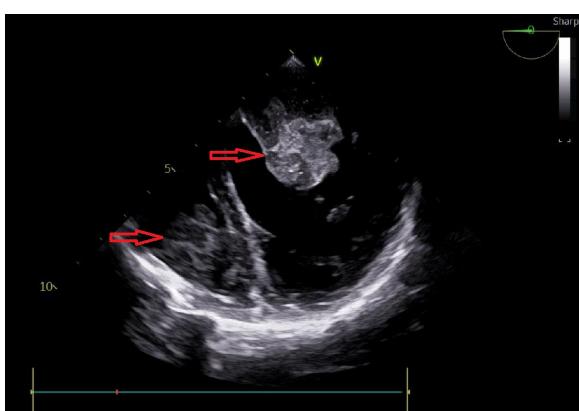


FIGURE 2
Transesophageal echocardiography, the mass shadow at the arrow is myxoma.

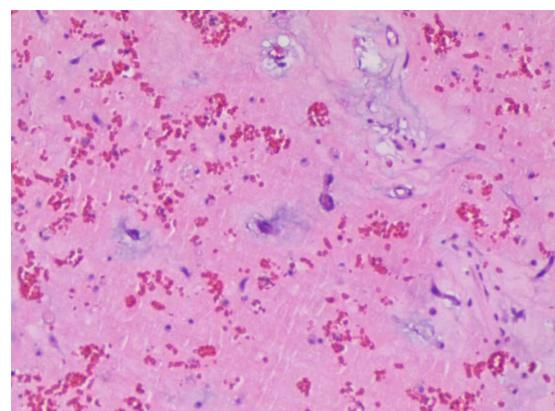


FIGURE 4
Sections under microscopic examination showed that the tumor cells are scattered in the myxoid matrix, star-shaped and polygonal, diffuse or intertwined into a network, and the focal area bleeds.

lasted a total of 76 min under cardiopulmonary bypass, with 49 min being cardiac function-blocked. Post-operation, the patient was transferred to the intensive care unit with endotracheal intubation, and regained consciousness 3 h later. The endotracheal tube was successfully removed on the second day after the operation.

The patient was discharged from the hospital 10 days after surgery in good spirits and the muscle strength of the limbs was normal. Despite the successful recovery in physical strength, there was an observed temporary decrease in the patient's calculation ability. However, during the 3-month follow-up, the patient exhibited no residual dysarthria or difficulty accessing words, and there were no recurrent ischemic events. Additionally, her calculation ability had returned to normal.

Discussion

As per our literature search, this is the first case report of multiple myxomas in left atrial and right ventricular inducing acute cerebral infarction in a young female patient. In this case, the patient was young and had cerebral infarction symptoms, which reminds doctors to pay attention to cardiogenic factor (4). In addition, the preoperative head magnetic resonance (MRI) of the patient showed that the areas of cerebral infarction were not confined to the same vascular area, which suggested that the stroke originated from the heart (5).

Cardiac MRI can provide unique information in the diagnosis of cardiac myxoma with robust tissue signature sequences (6). In this case, the patient suffered from acute cerebral infarction caused by cardiac myxomas. In order to prevent further aggravation of obstruction induced by cardiac myxomas, we performed emergency surgery as soon as possible to remove the risk of re-obstruction. The treatment of neurologic symptoms resulting from cardiac myxoma-induced infarction is a subject of debate, with no established guideline for stroke associated with myxoma (7). However, some scholars argue that immediate treatment of cardiac myxoma upon diagnosis is crucial for averting re-vascular obstruction, prioritizing it over the alleviation of neurological or other embolic symptoms in the brain (8).

Some scholars support the priority use of intravenous thrombolytic drugs after cerebral embolism, with recombinant tissue plasminogen activator considered safe and effective. However, postoperative pathology findings in successful thrombolysis cases revealed that the main component of the embolus was thrombosis. This finding may be attributed to the local vascular ischemic damage caused by myxoma embolus blocking cerebral blood vessels (9).

The choice of operation time window has also become a controversial topic. Wakako Fukuda et al. (10) conducted a study revealing that the occurrence of worsening brain complications decreased by 10% when surgery was performed more than 15 days after cerebral infarction. In the case reported by Yoshioka et al. (11), the patient's preoperative cranial MRI showed only

multiple small infarcts in the brain without any signs of cerebral hemorrhage. After emergency surgery, right occipital lobe cerebral hemorrhage was found, notably, the patient's preoperative test results, including a platelet count of only $1 \times 10^4/\text{mm}^3$ and a D-dimer level of 12.72 $\mu\text{g}/\text{ml}$, indicated severe disseminated intravascular coagulation (DIC). This case emphasizes the critical nature of preoperative coagulation function examination to avoid unforeseen complications during surgery.

In this case, after surgically removing the risk of cardiogenic obstruction, the neurological symptoms improved, and there were no new obstruction symptoms in other organs. Therefore, we believe it is crucial to promptly remove cardiac myxoma in non-DIC patients.

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

CT: Conceptualization, Formal Analysis, Writing – original draft. XG: Conceptualization, Writing – original draft. TC: Methodology, Writing – review & editing. JS: Investigation, Writing – review & editing. TZ: Visualization, Writing – review & editing. XZ: Formal Analysis, Investigation, Writing – review & editing. CR: Conceptualization, Funding acquisition, Writing – original draft.

Funding

The author(s) declare financial support was received for the research, authorship, and/or publication of this article.

Foundation of Yangzhou Science and Technology Planning (YZ2020076).

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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated

organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

References

1. Gošev I, Paić F, Durić Z, Gošev M, Ivčević S, Jakuš FB, et al. Cardiac myxoma the great imitators: comprehensive histopathological and molecular approach. *Int J Cardiol.* (2013) 164(1):7–20. doi: 10.1016/j.ijcard.2011.12.052
2. Wang JG, Li YJ, Liu H, Li NN, Zhao J, Xing XM. Clinicopathologic analysis of cardiac myxomas: seven years' experience with 61 patients. *J Thorac Dis.* (2012) 4(3):272–83. doi: 10.3978/j.issn.2072-1439.2012.05.07
3. Zhao S, Li H, Wu C, Pan Z, Wang G, Dai J. Surgical treatment of rare pediatric cardiac myxomas: 12 years clinical experience in a single institution. *BMC Cardiovasc Disord.* (2023) 23(1):219. doi: 10.1186/s12872-023-03255-2
4. Lm Silva M Jr, Albuquerque TE, Melo ES. Giant atrial myxoma leading to stroke. *Med J Aust.* (2021) 215(6):258–e1. doi: 10.5694/mja2.51231
5. Kelley RE, Kelley BP. Heart-brain relationship in stroke. *Biomedicines.* (2021) 9(12):1835. doi: 10.3390/biomedicines9121835
6. Beroukhim RS, Ghelani S, Ashwath R, Balasubramanian S, Biko DM, Buddhe S, et al. Accuracy of cardiac magnetic resonance imaging in diagnosing pediatric cardiac masses: a multicenter study. *JACC Cardiovasc Imaging.* (2022) 15(8):1391–405. doi: 10.1016/j.jcmg.2021.07.010
7. Al-Said Y, Al-Rached H, Baeesa S, Kurdi K, Zabani I, Hassan A. Emergency excision of cardiac myxoma and endovascular coiling of intracranial aneurysm after cerebral infarction. *Case Rep Neurol Med.* (2013) 2013:839270. doi: 10.1155/2013/839270
8. Isogai T, Yasunaga H, Matsui H, Tanaka H, Hisagi M, Fushimi K. Factors affecting in-hospital mortality and likelihood of undergoing surgical resection in patients with primary cardiac tumors. *J Cardiol.* (2017) 69(1):287–92. doi: 10.1016/j.jcc.2016.05.008
9. Acampi M, Guideri F, Tassi R, D'Andrea P, Marotta G, Lo Giudice G, et al. Thrombolytic treatment of cardiac myxoma-induced ischemic stroke: a review. *Curr Drug Saf.* (2014) 9(2):83–8. doi: 10.2174/157488630866140110123705
10. Eishi K, Kawazoe K, Kuriyama Y, Kitoh Y, Kawashima Y, Omae T. Surgical management of infective endocarditis associated with cerebral complications. Multi-center retrospective study in Japan. *Thorac Cardiovasc Surg.* (1995) 110(6):1745–55. doi: 10.1016/S0022-5223(95)70038-2
11. Yoshioka D, Takahashi T, Ishizaka T, Higuchi T. Successful surgical resection of infected left atrial myxoma in a case complicated with disseminated intravascular coagulation and multiple cerebral infarctions: case report. *Cardiothorac Surg.* (2011) 68(6). doi: 10.1186/1749-8090-6-68



OPEN ACCESS

EDITED BY

Giuseppe Gatti,
Azienda Sanitaria Universitaria Giuliano
Isontina, Italy

REVIEWED BY

Daniel P. Fudulu,
University of Bristol, United Kingdom
Davut Cekmecelioglu,
Cleveland Clinic, United States
Carlo Savini,
University of Bologna, Italy

*CORRESPONDENCE

Joscha Buech
✉ joscha.buech@med.uni-muenchen.de

RECEIVED 14 October 2023

ACCEPTED 22 January 2024

PUBLISHED 06 February 2024

CITATION

Buech J, Radner C, Fabry T, Rutkowski S, Hagl C, Peterss S and Pichlmaier MA (2024) Case Report: Incidental finding of an atresia of the inferior vena cava—a challenge for cardiac surgery. *Front. Cardiovasc. Med.* 11:1321685. doi: 10.3389/fcvm.2024.1321685

COPYRIGHT

© 2024 Buech, Radner, Fabry, Rutkowski, Hagl, Peterss and Pichlmaier. This is an open-access article distributed under the terms of the [Creative Commons Attribution License \(CC BY\)](#). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

Case Report: Incidental finding of an atresia of the inferior vena cava—a challenge for cardiac surgery

Joscha Buech^{1,2,3*}, Caroline Radner^{1,2,3}, Thomas Fabry^{1,3},
Simon Rutkowski^{1,3}, Christian Hagl^{1,2,3}, Sven Peterss^{1,3} and
Maximilian A. Pichlmaier^{1,3}

¹Department of Cardiac Surgery, LMU University Hospital, Munich, Germany, ²German Centre for Cardiovascular Research (DZHK), Partner Site Munich Heart Alliance, Munich, Germany, ³University Aortic Center Munich (LMU), LMU University Hospital, Munich, Germany

Inferior vena cava atresia is a rare and usually asymptomatic condition. However, when these patients undergo cardiac surgery, it can present an unexpected and challenging situation for the surgeon. Specifically, adequate venous drainage during cardiopulmonary bypass (CPB) is a critical issue here and may require an extension of cannulation strategies. Adequate preoperative diagnostics, ideally with imaging modalities such as CT angiography or MRI, are required for optimal surgical planning. Here, we describe a rare case of thoracic ascending aortic aneurysm with concomitant inferior vena cava atresia that was successfully operated on. With adequate preoperative planning, we were able to perform an operation without unforeseen complications with standard initialization of CPB.

KEYWORDS

aortic surgery, thoracic aorta aneurysm, anatomic variant, vena cava inferior agenesis, computer tomograph, preoperating planning

Introduction

Continuity of the azygos vein with an interrupted or totally absent inferior vena cava (IVC) is a rare congenital anomaly affecting 0.3% of otherwise healthy individuals (1). IVC interruption may be found in up to 2% of patients presenting with other cardiovascular defects such as dextrocardia, atrial septal defect, atrioventricular canal, and pulmonary artery stenosis (2). In everyday life, patients with an interruption of the IVC are often asymptomatic but have an increased risk of venous thrombosis (3, 4). If cardiac surgery with CPB is required, strategies are needed to ensure unobstructed venous drainage in these patients, as venous return may be compressed in the supine position on the operating table due to the lack of IVC. Therefore additional venous cannulation e.g., of the femoral vein may be required to allow the CPB to perform the a full calculated cardiac output. Furthermore, if unknown to the surgeon, agenesis of the IVC may lead to fatal injury to the hepatic veins during the attempt to cannulate the non-existent IVC via the right atrium.

Here, we describe the case of a patient with incidental finding of an agenesis of the IVC and continuity of the azygos vein who underwent aortic surgery.

Case presentation

A 54-year-old woman presented with severe aortic valve stenosis and an ascending aortic aneurysm. Echocardiography revealed a maximum gradient across the aortic valve of

58 mmHg, a calculated valve opening area of 0.7 cm^2 , a bicuspid valve morphology, and ventricular hypertrophy. Left ventricular function was normal. CT angiography confirmed an aneurysm of the ascending aorta of $46 \times 45 \text{ mm}$. Additionally, an azygous continuity with complete agenesis of the inferior vena cava was found incidentally in routine CT angiography. Both iliac veins were found to directly drain into the corresponding paravertebral vein and then merged with the left and right renal veins to form a large azygous vein (Figure 1).

An elective replacement of the aortic root, ascending aorta and proximal aortic arch was scheduled. During induction of anaesthesia, an additional distal venous pressure measurement was established via a femoral vein catheter. Initial measurement showed no difference to the central venous pressure measured in the internal jugular vein. Intraoperatively, this monitoring strategy served to ensure adequate drainage of the lower body throughout CPB.

Following median sternotomy and pericardiotomy, cannulation of the ascending aorta as well as the superior vena cava via the right atrium were performed, the latter using a regular two-stage cannula directed cranialwards (Figure 2). CPB was initiated and proved satisfactory considering venous drainage and full relief of the heart. Additional femoral venous cannulation was not required. For cardiac arrest, ice-cold ($5-8^\circ\text{C}$) crystalloid solution was used. Additionally topical cooling with cold water was performed. The

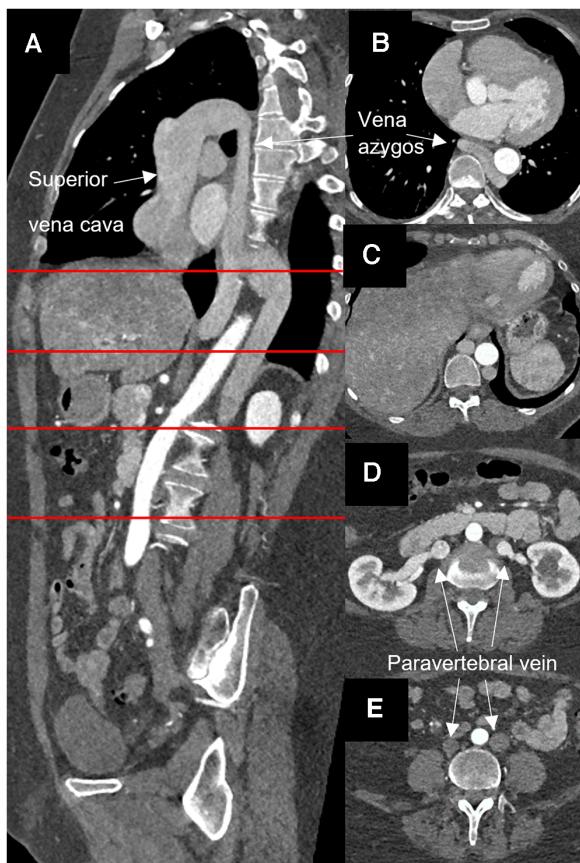


FIGURE 1
Computed angiography showing atresia of the inferior vena cava and azygous continuity. (A) Sagittal layer, (B–E) horizontal layer at the sites marked in red.

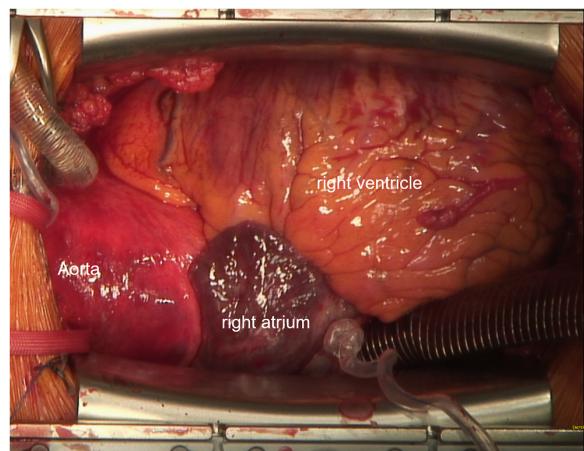


FIGURE 2
Intraoperative cannulation. Venous cannulation with a 2-stage cannula into the superior vena cava is shown.

aortic valve, aortic root, and ascending aorta were replaced using a mechanical 23 mm conduit. The hemiarch was replaced with a 26 mm tube under hypothermic circulatory arrest (25 min) with selective antegrade cerebral perfusion (SACP). Therefore, after reaching a body temperature of 25°C , SACP was initiated by open direct cannulation of the innominate artery and the left common carotid artery. The left subclavian artery was blocked with a balloon catheter. The total duration of the cardiopulmonary bypass was 4 h and 26 min, and aortic clamp time was 2 h and 46 min.

The patient was transferred to the intensive care unit. Weaning from ventilation took place 12 h after surgery, and the patient was transferred to the intermediate care ward. During the hospital stay, the patient experienced a pericardial effusion requiring re-sternotomy. The remaining hospital stay was uneventful, and the patient was discharged in good physical condition. The predischarge CT angiography confirmed a satisfactory early postoperative result.

Discussion

Atresia of the IVC with azygous continuity is a challenging condition in case cardiac surgery is needed. Recently, two cases of cardiac surgery with an aberrant IVC have been described. Cetinkaya et al. reported a case of a 61-year-old man with mitral and tricuspid valve replacement and an abnormal intraoperative IVC finding requiring conversion from a minimally invasive approach to a full sternotomy (5). Knol et al. describe a similar case of a 57-year-old man with mitral and tricuspid insufficiency where a full sternotomy was also required after initially starting with a minimally invasive approach for valve reconstruction (6). In both cases, additional cannulation of the femoral vein was required for adequate venous drainage. In this situation, a hybrid room for positioning the venous cannula with angiographic support could be useful as transesophageal echocardiography may not provide sufficient visibility to check the correct position. This should be considered in the preoperative planning. In our case, a central two-stage venous cannula directed

upwards rather than downwards proved sufficient. Monitoring of the venous pressure in the iliac veins provided the critical information about sufficient venous drainage throughout the operation. This additional monitoring was also used for postoperative fluid management and catecholamine dosing in the intensive care unit.

This case underlines the importance of thorough preoperative diagnostics in elective cardiac surgery in order to be able to recognize and prepare different strategies even for such a rare anatomical variant. Non-invasive preoperative diagnostics such as computed tomography or magnetic resonance imaging becoming increasingly important in preoperative diagnostics and are suited to depict also the IVC but are not yet standard care in cardiac surgery (7, 8). The chest x-ray may show unspecific signs like a widened mediastinum with an enlarged azygous arch (9). In infants, ultrasound with the specific “double sign” may be an alternative (10). In adults, transesophageal ultrasound showing the interruption of back flow can be a diagnostic sign (11).

Conclusion

We hereby describe a rare case of IVC atresia with azygous continuity in a patient undergoing thoracic aortic surgery. It could be demonstrated that, with proper preoperative diagnostics and planning, this challenging condition may even be addressed without the need for a strategy change during the operation.

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 Ethics Committee of the Medical Faculty of LMU Munich. The studies

References

1. Bass JE, Redwine MD, Kramer LA, Huynh PT, Harris JH. Spectrum of congenital anomalies of the inferior vena cava: cross-sectional imaging findings. *Radiographics*. (2000) 20(3):639–52. doi: 10.1148/radiographics.20.3.g00ma09639
2. Oliveira JD, Martins I. Congenital systemic venous return anomalies to the right atrium review. *Insights Imaging*. (2019) 10(1):115. doi: 10.1186/s13244-019-0802-y
3. de la Morena-Barrio ME, Gindels R, Bravo-Pérez C, Ilonczai P, Zuazu I, Speker M, et al. High penetrance of inferior vena cava system atresia in severe thrombophilia caused by homozygous antithrombin budapest 3 variant: description of a new syndrome. *Am J Hematol.* (2021) 96(11):1363–73. doi: 10.1002/ajh.26304
4. Mabud TS, Sailer AM, Swee JKY, Tamboli M, Arendt VA, Jeon GS, et al. Inferior vena cava atresia: characterisation of risk factors, treatment, and outcomes. *Cardiovasc Intervent Radiol.* (2020) 43(1):37–45. doi: 10.1007/s00270-019-02353-z
5. Cetinkaya A, Bramlage P, Schönburg M, Richter M. Atresia of the inferior vena cava in a patient undergoing mitral and tricuspid valve surgery. *Interact Cardiovasc Thorac Surg.* (2019) 28(2):324–6. doi: 10.1093/icvts/ivy240
6. Knol WG, Oei FB, Budde RPJ, Ter Horst M. A case report of an interrupted inferior vena cava and azygous continuation: implications for preoperative screening in minimally invasive cardiac surgery. *Eur Heart J Case Rep.* (2021) 5(8).
7. Koc Z, Oguzkurt L. Interruption or congenital stenosis of the inferior vena cava: prevalence, imaging, and clinical findings. *Eur J Radiol.* (2007) 62(2):257–66. doi: 10.1016/j.ejrad.2006.11.028
8. Kulkarni S, Szeto WY, Jha S. Preoperative computed tomography in the adult cardiac surgery patient. *Curr Probl Diagn Radiol.* (2022) 51(1):121–9. doi: 10.1067/j.cpradiol.2020.10.008
9. Saito T, Watanabe M, Kojima T, Matsumura T, Fujita H, Kiyosue A, et al. Successful blood sampling through azygous continuation with interrupted inferior vena cava. A case report and review of the literature. *Int Heart J.* (2011) 52(5):327–3. doi: 10.1536/ihj.52.327
10. Sheley RC, Nyberg DA, Kapur R. Azygous continuation of the interrupted inferior vena cava: a clue to prenatal diagnosis of the cardiosplenic syndromes. *J Ultrasound Med.* (1995) 14(5):381–7. doi: 10.7863/jum.1995.14.5.381
11. Pantin EJ, Naftalovich R, Denny J. Echocardiographic identification of an interrupted inferior vena cava with dilated azygous vein during coronary artery bypass graft surgery. *Anesth Analg.* (2016) 122(2):358–60. doi: 10.1213/ANE.0000000000001075



OPEN ACCESS

EDITED BY

Enyi Shi,
China Medical University, China

REVIEWED BY

Karthik Seetharam,
West Virginia State University, United States
Massimo Baudo,
Lankenau Institute for Medical Research,
United States
Mohamed Rahouma,
Weill Cornell Medical Center, United States

*CORRESPONDENCE

Jiahai Shi
✉ happysjh167@163.com

RECEIVED 18 October 2023

ACCEPTED 05 February 2024
PUBLISHED 14 February 2024

CITATION

Xiao W, Qin J, Feng J, Jiang F, Chen X, Cao X, Xue Q and Shi J (2024) Case Report: Giant left atrial cystic tumor: myxoma or intracardiac blood cyst? *Front. Cardiovasc. Med.* 11:1323890. doi: 10.3389/fcvm.2024.1323890

COPYRIGHT

© 2024 Xiao, Qin, Feng, Jiang, Chen, Cao, Xue and Shi. This is an open-access article distributed under the terms of the [Creative Commons Attribution License \(CC BY\)](#). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

Case Report: Giant left atrial cystic tumor: myxoma or intracardiac blood cyst?

Weizhang Xiao¹, Jing Qin², Jia Feng³, Feng Jiang², Xinming Chen¹, Xiang Cao¹, Qun Xue¹ and Jiahai Shi^{1*}

¹Department of Cardiothoracic Surgery, Affiliated Hospital and Medical School of Nantong University, Nantong, China, ²Department of Echocardiography, Affiliated Hospital and Medical School of Nantong University, Nantong, China, ³Department of Pathology, Affiliated Hospital and Medical School of Nantong University, Nantong, China

Background: Primary cardiac tumors are uncommon, with the majority being benign myxomas. Cystic myxoma, a particularly rare type of benign cardiac tumor, demands cautious differential diagnosis from other cardiac tumors.

Case summary: A 43-year-old male patient presenting with intermittent dyspnea was referred to our department for surgical evaluation. Transthoracic echocardiography (TTE) and transesophageal echocardiography (TEE) unveiled an intra-left atrial cyst, which was subsequently found to be blood-filled during a video-assisted microinvasive heart surgery. Pathological examination depicted a cyst wall filled with small stellate and fat spindle cells, along with a mucoid matrix, indicating a diagnosis of cystic myxoma.

Conclusions: We herein presented a rare case of an adult patient with cystic myxoma, initially misdiagnosed as an intracardiac blood cyst (CBC) prior to surgery, and ultimately verified via pathological findings.

KEYWORDS

case report, cardiac tumor, myxoma, intracardiac blood cyst, cardiac surgery

Introduction

Primary cardiac tumors are rare, with an incidence of 0.002% to 0.3% and a prevalence of 0.001% to 0.03%. Over half of these tumors are myxomas (1). Most myxomas occur in the left atrium (75% to 80%), with fewer cases found in the right atrium (10% to 20%) (2). Typically, these tumors are solid, round, or polypoid in shape and attached to the interatrial septum, lacking a cystic structure. However, cystic myxomas are exceedingly rare (3–8). Intracardiac blood cysts are another unusual cardiac tumor, mainly seen in fetuses or infants under six months of age and rarely reported in adults. Here, we present an uncommon case of a left atrial cystic myxoma that was initially suspected as a CBC before surgery and ultimately diagnosed as a cystic myxoma.

Patient information

A 43-year-old male patient presented with intermittent dyspnea during sleep for 20 days and underwent TTE at a local hospital, which revealed a left atrial myxoma. He was referred to our hospital for surgical evaluation and was further directed to the cardiothoracic surgery department. The patient denied any history of hypertension,

diabetes mellitus, and tobacco use. Upon physical examination, a moderate diastolic murmur was auscultated at the cardiac apex area. Electrocardiography showed a sinus rhythm with a heart rate of 63 beats per minute. All initial laboratory tests were within normal limits, except for a mild elevation in B-type natriuretic peptide (108 pg/L). Additionally, hemoglobin was 143 g/L, albumin was 38.5 g/L, aspartate aminotransferase was 13 U/L, alanine aminotransferase was 12 U/L, erythrocyte sedimentation rate was 2 mm/hour, and Troponin was 0.011 µg/L. TTE demonstrated an almost echoless mass measuring 4.6 × 4.4 cm with well-defined margins in the left atrium, which was attached to the atrial septum with a narrow base (Figures 1A,B). TEE showed a large cystic mass in the left atrium that protruding into the mitral valve during diastole, causing mild mitral stenosis (Supplementary Video S1). A cardiac CT scan

displayed a giant tumor located in the left atrium with heterogeneous density following enhancement, suggesting the possibility of a myxoma (Figures 1C,D).

The patient underwent a video-assisted thoracic surgery using the Da Vinci surgical system (Intuitive Surgical Inc., Sunnyvale, CA, USA). After double-lumen intubation, cardiopulmonary bypass (CPB) was established by femoral artery and vein cannulation, and a right-sided incision was made to access the chest cavity. During the surgery, a giant parenchymal cyst was found in the left atrium with a pedicle attached to the atrial septum, which was intact (Figure 2A). We attempted to completely remove this lesion. Unfortunately, the cyst accidentally ruptured during resection, causing blood to flow out and resulting in its collapse. Nevertheless, the tumor was completely removed without causing any damage to the

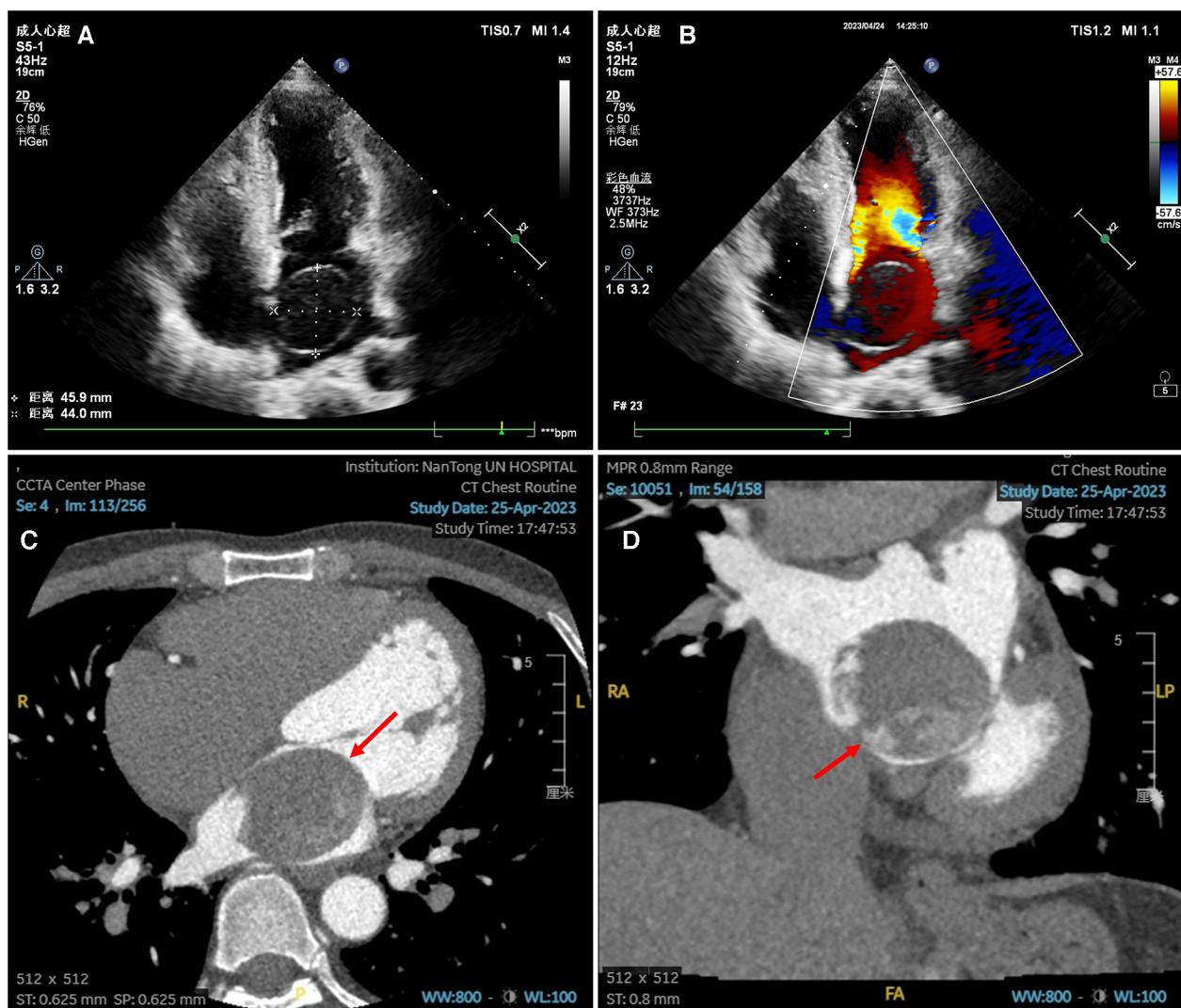


FIGURE 1

Preoperative echocardiography and enhanced cardiac CT scan images. (A–B) Transthoracic echocardiography showed an echoless tumor measuring 4.6 × 4.4 cm in the left atrium, with clear boundary, a small base attached to the atrial septum, and no blood signal in the tumor. (C–D) Enhanced cardiac CT scan in transverse plane (C) and coronal plane (D) showed a giant tumor in the left atrium with heterogeneous density after enhancement.

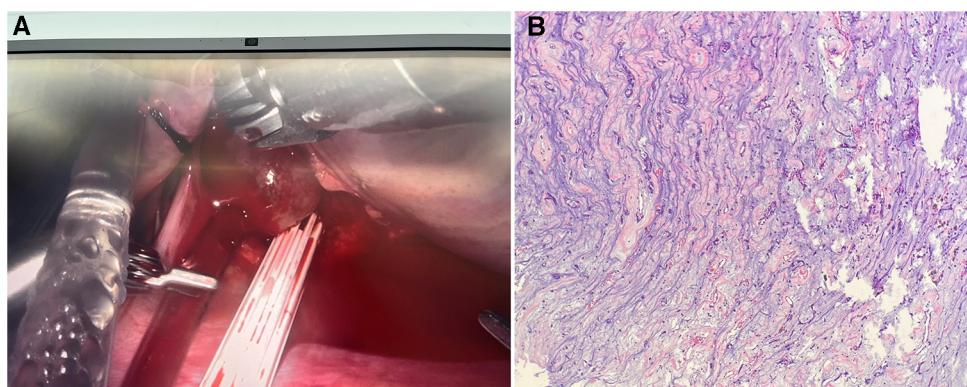


FIGURE 2

Intraoperative and pathological images. (A) Intraoperative view revealed a parenchymal blood-filled cyst with smooth surface attached to the atrial septum with a pedicle. (B) Pathological examination of the cyst (hematoxylin and eosin stain, $\times 4$) showed spindle cells with eosinophilic cytoplasm surrounded by mucoid matrix.

adjacent tissue. The total operation time was 2 h and 14 min, the CPB time was 1 h and 18 min, and the cross-clamp time was 35 min. Pathological examination revealed a smooth cyst wall with a thickness of 0.1–0.2 cm. The cyst wall consisted of small stellate and fat spindle cells with round, oval, or elongated nuclei and eosinophilic cytoplasm, surrounded by a mucoid matrix. The tumor cells were arranged in a linear pattern and oriented towards the blood vessels, with no cytological atypia present (Figure 2B). The postoperative recovery process was uneventful, and the patient was transferred from the intensive care unit to the general ward on the second day after surgery, and discharged nine days later, with normal mitral valve and left ventricular functions confirmed through echocardiography before discharge. During the follow-up period, there was no recurrence of myxoma.

Discussion

As a very rare tumor, the pathological type of cardiac mass is closely related to its location. For instance, myxoma can always be seen in the left atrium, lipoma is more common in the right atrium or left ventricle, and fibroma and rhabdomyoma are more common in the ventricles (9). While most diagnoses of cardiac tumors are rendered through imaging techniques, some of them may not exhibit characteristic imaging features that suggest their pathological type, making the diagnosis challenging. Therefore, myocardial biopsy plays an important role in the diagnosis of cardiac tumors. Currently, computerized tomography (CT), TEE, or intracardiac echocardiography are commonly used to guide myocardial biopsy (10). Among them, CT-guided biopsy has been proven to be safe for pericardial or superficial intermural tumors (11). For intracardiac masses, myocardial biopsy is most performed via the venous route, including internal jugular or femoral vein, and for left ventricular lesions, biopsy can also be performed by transseptal puncture or directly through peripheral

arteries (12). Considering the high risk of myocardial biopsy including vasovagal reaction, pericardial tamponade, arrhythmias, ventricular perforation, and vascular injury (13), it is generally utilized for tumors in the right cardiac system (14). Additionally, myocardial biopsy is considered contraindicated in the following situations: (1) cardiac tumors without a safe puncture path; (2) the surface of mass is surrounded by large tortuous blood vessels; (3) preoperative imaging showing significant necrosis within the lesion or severe coagulation dysfunction; (4) severe dyspnea or agitation; and (5) friable masses with a high embolic potential, such as left ventricular tumors or typical cardiac myxomas (11).

As the most common cardiac tumor, the clinical features of cardiac myxoma depend on the location and size of tumor. Dyspnea is the most frequent symptom of left atrial myxoma, which is a characteristic manifestation of mitral valve dysfunction leading to left-sided heart failure. Conversely, myxoma in the right atrium may result in symptoms related to right-sided heart failure. Given that the majority of cardiac myxomas exhibit a non-cystic structure, embolism should be of particular concern, with approximately half of all cases presenting embolism-related events, including strokes, retinal artery emboli, and limb embolism (15).

Cystic myxoma is a relatively rare condition that is typically identified incidentally via TTE. Through searching PubMed, only a few literatures were found to report the existence of cystic myxoma as shown in Table 1. Reports have indicated the presence of feeding arteries into the tumor, originating from either the left or right coronary artery, potentially leading to coronary steal phenomenon such as angina (5, 8). We hypothesize that hemorrhage within the tumor contributes to the formation of a cystic mass filled with blood. Moreover, the stability of this blood-filled cyst requires a sufficient drainage hole for outlet flow. Diagnosing cystic myxoma requires caution, as the differential diagnosis includes several other conditions, such as CBC, hemangioma, angiosarcoma, hydatid cyst, thrombus, and metastatic tumors (8, 21).

TABLE 1 Reported cases of cystic cardiac myxoma in the literature till January 2024.

Numbers	Author, year	Age	Gender	Chief complaint	Location	Size
1	Okuri H. (16)	54	Male	Dyspnea	Right atrium	69 × 44 mm
2	Lee KT. (17)	72	Female	Dyspnea	Left atrium	30 × 30 mm
3	Benezet-Mazuecos J. (18)	59	Female	Dyspnea	Left atrium	55 × 30 mm
4	Park J. (19)	71	Male	Dyspnea	Left atrium	53 × 32 mm
5	Acikel S. (5)	38	Female	Dyspnea, angina	Left atrium	75 × 50 mm
6	Park, KJ. (6)	65	Female	Fever, malaise	Left atrium	24 × 23 mm
7	Toprak, C. (7)	47	Male	Dyspnea	Left atrium	58 × 38 mm
8	Liao JM. (20)	35	Female	Dyspnea	Left atrium	70 × 50 mm
9	Watanabe H. (21)	75	Male	Dyspnea	Left atrium	54 × 39 mm
10	Shabestari MM. (22)	69	Female	Dyspnea, chest pain	Left atrium	26 × 25 mm
11	Xie, X. (4)	62	Female	Dyspnea	Left atrium	35 × 30 mm
12	Ntinopoulos V. (3)	63	Female	Dyspnea	Left atrium	29 × 22 mm
13	Suzuki T. (23)	44	Male	Dyspnea	Left atrium	N/A
14	Azad S. (24)	11	Male	Chest and throat pain	Left ventricle	39 × 26 mm
15	Futami S. (8)	73	Male	N/A	Left atrium	32 × 24 mm

CBC, first documented in 1,844, remains a relatively uncommon benign cardiac tumor, primarily discovered during autopsy and rarely reported in adults (25). Many patients with CBC exhibit no symptoms and are often diagnosed incidentally during routine echocardiography. Nevertheless, CBCs positioned in the semilunar or atrioventricular valves, as is most commonly the case, may induce valve dysfunction.

In the present case, given the smooth and thin morphology of the cyst wall, a CBC was strongly suspected prior to surgery. This suspicion was further reinforced by the intraoperative observations, which revealed a blood-filled, balloon-like cyst. Although an enhanced cardiac CT scan failed to demonstrate any feeding arteries within the tumor, histopathological examination was crucial for arriving at a definitive diagnosis. The result revealed a cyst wall exhibiting myxoma manifestations and lacking endothelial structure, confirmatively indicating a cystic myxoma.

Conclusion

In this case report, we present an extremely rare case of a cystic left atrial myxoma. Preoperative echocardiography of the patient revealed a thin-walled cyst, which was further confirmed during surgery. Additionally, the cystic fluid was found to be bloody, leading us to initially suspect it as a CBC. However, upon the histopathological examination of the tissue, it was conclusively diagnosed as a cystic myxoma rather than CBC due to the absence of endothelial structure in the cyst wall and the presence of distinctive histological features of myxoma.

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 Institutional Review Board (IRB) and ethics committee of the Affiliated Hospital of Nantong 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

WX: Conceptualization, Methodology, Writing – original draft. JQ: Data curation, Software, Writing – original draft. JF: Data curation, Software, Writing – review & editing. FJ: Data curation, Software, Writing – review & editing. XCh: Data curation, Methodology, Writing – review & editing. XCa: Methodology, Writing – review & editing. QX: Conceptualization, Supervision, Writing – review & editing. JS: Conceptualization, Funding acquisition, Supervision, 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 (82370253), Jiangsu Provincial Research Hospital (YJXYY202204), Innovation Team Project of Affiliated Hospital of Nantong University (XNBHCX31773).

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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated

organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

Supplementary material

The Supplementary Material for this article can be found online at: <https://www.frontiersin.org/articles/10.3389/fcvm.2024.1323890/full#supplementary-material>

References

- Rahouma M, Arisha MJ, Elmously A, El-Sayed Ahmed MM, Spadaccio C, Mehta K, et al. Cardiac tumors prevalence and mortality: a systematic review and meta-analysis. *Int J Surg.* (2020) 76:178–89. doi: 10.1016/j.ijsu.2020.02.039
- Layton S, Ripley DP, Bellenger NG. Left atrial myxoma. *Br Med J.* (2013) 347:f4430. doi: 10.1136/bmj.f4430
- Ntinopoulos V, Dushaj S, Brugnetti D, Rings L, Loeblein H, Dzemali O. Left atrial myxoma: unusual presentation as a cystic tumor. *J Card Surg.* (2020) 35:511–3. doi: 10.1111/jocs.14401
- Xie X, Bai J. Left atrial myxoma presenting as a cystic mass. *J Card Surg.* (2017) 32:694–5. doi: 10.1111/jocs.13224
- Acikel S, Aksoy MM, Kilic H, Karapinar K, Oguz AS, Aydin H, et al. Cystic and hemorrhagic giant left atrial myxoma in a patient presenting with exertional angina and dyspnea. *Cardiovasc Pathol.* (2012) 21:e15–8. doi: 10.1016/j.carpath.2011.01.006
- Park KJ, Woo JS, Park JY. Left atrial myxoma presenting with unusual cystic form. *Korean J Thorac Cardiovasc Surg.* (2013) 46:362–4. doi: 10.5090/kjtcs.2013.46.5.362
- Toprak C, Kahveci G, Tabakci MM, Acar G, Emiroglu MY. Unusual image of a cystic atrial myxoma: mass in mass appearance in the left atrium. *Herz.* (2015) 40:259–60. doi: 10.1007/s00059-013-3923-y
- Futami S, Hieda M, Fukata M, Shiose A. A rare case of cardiac myxoma with light bulb-like cystic morphology: a case report. *Eur Heart J Case Rep.* (2023) 7:ytad331. doi: 10.1093/ehjcr/ytad331
- Seferović PM, Tsutsui H, McNamara DM, Ristić AD, Basso C, Bozkurt B, et al. Heart failure association of the ESC, heart failure society of America and Japanese heart failure society position statement on endomyocardial biopsy. *Eur J Heart Fail.* (2021) 23:854–71. doi: 10.1002/ejhf.2190
- Naruse G, Kawasaki M, Yanase K, Tanaka T. Primary angiosarcoma in the right atrium diagnosed by a cardiac tumor biopsy using intracardiac echocardiography. *J Med Ultrasound.* (2020) 28:120–2. doi: 10.4103/jmu.JMU_93_19
- Xie Y, Hong ZL, Zhao YC, Chen S, Lin YC, Wu SS. Percutaneous ultrasound-guided core needle biopsy for the diagnosis of cardiac tumors: optimizing the treatment strategy for patients with intermural and pericardial cardiac tumors. *Front Oncol.* (2022) 12:931081. doi: 10.3389/fonc.2022.931081
- Veinot JP. Diagnostic endomyocardial biopsy pathology-general biopsy considerations, and its use for myocarditis and cardiomyopathy: a review. *Can J Cardiol.* (2002) 18:55–65. PMID: 11826329
- Oliveira GH, Al-Kindi SG, Hoimes C, Park SJ. Characteristics and survival of malignant cardiac tumors: a 40-year analysis of >500 patients. *Circulation.* (2015) 132:2395–402. doi: 10.1161/CIRCULATIONAHA.115.016418
- Veinot JP. Endomyocardial biopsy-when and how. *Cardiovasc Pathol.* (2011) 20:291–6. doi: 10.1016/j.carpath.2010.08.005
- Butany J, Nair V, Naseemuddin A, Nair GM, Catton C, Yau T. Cardiac tumours: diagnosis and management. *Lancet Oncol.* (2005) 6:219–28. doi: 10.1016/S1470-2045(05)70093-0
- Okuri H, Shimizu M, Yokoyama K, Kawada H, Irisawa A, Kikawada R. A case of right atrial myxoma: M-mode and pulsed-doppler echocardiographic findings before and after operation. *Kokyū to Junkan.* (1993) 41:397–401. PMID: 8516580
- Lee KT, Lai WT, Yen HW, Voon WC, Hwang CH, Lu YH, et al. Cystic left atrium myxoma—a rare case report. *Kaohsiung J Med Sci.* (2001) 17:579–81. PMID: 11852466
- Benezet-Mazuecos J, Marcos-Alberca P, Farre J, Manzarbeitia F, Rabago R, Rey M. Multicystic/cavitated giant left atrial myxomas: a matter of technology. *Eur J Echocardiogr.* (2008) 9:101–2. doi: 10.1016/j.euje.2007.03.040
- Park J, Song JM, Shin E, Jung SH, Kim DH, Kang DH, et al. Cystic cardiac mass in the left atrium: hemorrhage in myxoma. *Circulation.* (2011) 123:e368–9. doi: 10.1161/CIRCULATIONAHA.110.004655
- Liao JM, Nasseri F, Nachiappan AC, Kuban J, Cheong BY. Left atrial myxoma presenting as a cystic mass. *Tex Heart Inst J.* (2013) 40:358–9. PMID: 23914040
- Watanabe H, Nara I, Yamaura G, Iino K, Iino T, Shimbo M, et al. Blood balloon induced by an atrial myxoma in the heart. *Circulation.* (2014) 130:2351–3. doi: 10.1161/CIRCULATIONAHA.114.010732
- Shabestari MM, Fazlinezhad A, Moravvej Z, Tashnizi MA, Azari A, Bigdelu L. A case of left atrial myxoma with unusual tumor vascularity. *Asian Cardiovasc Thorac Ann.* (2015) 23:458–60. doi: 10.1177/0218492313513776
- Suzuki T, Hata M, Yamaya K, Saitou T, Haba F, Matsuno M, et al. Cystic myxoma which obstructed the mitral valve orifice; report of a case. *Kyobu Geka.* (2020) 73:380–3. PMID: 32398397
- Azad S, Dutta N, Roy Chowdhuri K, Ramman TR, Chandra N, Radhakrishnan S, et al. Atypical left ventricular myxoma: unusual echocardiographic and histopathological features. *World J Pediatr Congenit Heart Surg.* (2020) 11:NP129–129NP131. doi: 10.1177/2150135117742626
- Halim J, van Schaagen FR, Riezebos RK, Lalezari S. Giant intracardiac blood cyst: assessing the relationship between its formation and previous cardiac surgery. *Neth Heart J.* (2015) 23:392–4. doi: 10.1007/s12471-015-0707-4



OPEN ACCESS

EDITED BY

Giuseppe Gatti,
Azienda Sanitaria Universitaria Giuliano
Isontina, Italy

REVIEWED BY

Abimbola Faloye,
Emory University, United States
Philemon Gukop,
St George's University Hospitals NHS
Foundation Trust, United Kingdom

*CORRESPONDENCE

Hassan Tatari
✉ tatari@sbmu.ac.ir

RECEIVED 29 November 2023

ACCEPTED 22 January 2024

PUBLISHED 23 February 2024

CITATION

Khorgami M, Khalaj F, Gholampour M and Tatari H (2024) Missile embolism from pulmonary vein to left ventricle: report of a case.
Front. Cardiovasc. Med. 11:1342146.
doi: 10.3389/fcvm.2024.1342146

COPYRIGHT

© 2024 Khorgami, Khalaj, Gholampour and Tatari. This is an open-access article distributed under the terms of the [Creative Commons Attribution License \(CC BY\)](#). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

Missile embolism from pulmonary vein to left ventricle: report of a case

Mohammadrafe Khorgami¹, Fattaneh Khalaj²,
Maziar Gholampour¹ and Hassan Tatari^{1*}

¹Rajaie Cardiovascular Medical and Research Center, Iran University of Medical Sciences, Tehran, Iran,

²Liver and Pancreatobiliary Diseases Research Center, Digestive Diseases Research Institute, Tehran University of Medical Sciences, Tehran, Iran

Missile embolization is rare in penetrating trauma, occurring in 0.3% of cases. Bullet embolism into the left ventricle is less frequent, with few instances described in the literature. This paper describes an instance of left ventricular bullet embolism from the pulmonary venous system following gunshot chest trauma. A 7-year-old boy sustained a gunshot wound to his chest during an assault accident. Despite thoracic pain, he remained conscious and exhibited vital signs. A CXR and CT scan revealed a bullet in the left mediastinum. A left thoracotomy was performed to remove blood and clots from the pericardium. The patient was sent to a tertiary referral hospital for further investigation. The patient underwent elective surgery to remove the foreign body from inside the heart. The procedure involved a partial thymectomy and pericardial opening, and the patient was released from medical care after 14 days. After 6 months, there were no signs or symptoms of cardiothoracic infection or evidence of mitral valve regurgitation in echocardiography.

KEYWORDS

missile embolization, gunshot, left ventricle, pulmonary vein (PV), foreign body

1 Introduction

Missile embolization is a rare occurrence in penetrating trauma, where an object enters the bloodstream and travels to another part of the vascular system. In fact, according to a review of over 7,500 casualties from the Vietnam War, bullet embolization occurred only in 0.3% of cases (1). Although reports suggest emboli rarely occur in the pulmonary vasculature and right heart, bullet embolism into the left ventricle following penetrating chest injuries is considerably less frequent with few instances described in the literature (2–7). Due to a lack of extensive experience at any single institution, there are several controversies surrounding appropriate diagnostic and therapeutic approaches for managing such cases. This paper describes an instance of left ventricular bullet embolism from the pulmonary venous system following gunshot chest trauma.

2 Case description

The presented case report describes a 7-year-old boy without history of other medical concerns who was brought to the emergency unit after sustaining a single gunshot wound to the right side of his chest, resulting from an assault accident while on his way to school. There was no exit wound. Despite complaining of thoracic pain, the boy remained

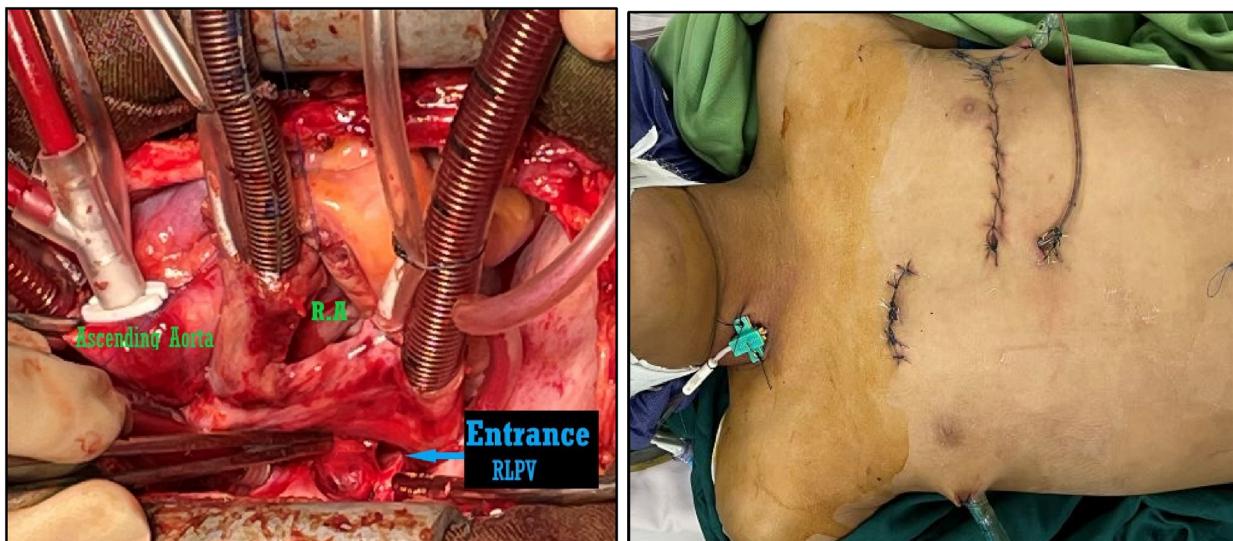


FIGURES 1, 2

According to the CXR, a bullet of 5.5 mm in length was found in the anteroposterior of left part of the mediastinum, gunshot.

conscious and exhibited vital signs, including a blood pressure of 116/79 mm Hg, a pulse rate of 118 beats per min, and a respiratory rate of 38/min. After examination by a general surgeon, a chest tube was placed in the damaged thorax (right), and a small amount of blood and air were released. Further examination following the relative stability of hemodynamics revealed an entrance wound near the right side of the sternum at the level of the third intercostal space. There was no marked subcutaneous emphysema or jugular venous distention. His initial hematocrit was 32%. Arterial blood gases did not detect hypoxemia or acidosis, and an electrocardiogram showed normal

sinus rhythm. Upon conducting a chest radiograph (CXR) and computerized tomography (CT) scan, it was determined a bullet (5.5 mm long) was in the left part of the mediastinum (Figures 1, 2). Therefore, a pleuropericardial window is installed through the left thoracotomy, which removes some blood and clots from the pericardium. After completing these procedures and receiving intravenous fluids, the patient's vital signs became completely stable, and he was referred to the local hospital with a cardiologist for further investigation, where echocardiography was performed and placing the bullet in the left ventricle was confirmed. Considering the risk of embolization, the patient is



FIGURES 3, 4

The entrance location of the bullet was discovered after right atriotomy and septostomy in the anterosuperior portion of the lower right pulmonary vein, and the bullet that had been lodged in the posterior chordal lobe of the mitral valve was removed and chest wall was repaired.

sent to the tertiary referral hospital. Transesophageal echocardiography (TEE) showed that the right atrium and ventricle were functioning normally, with a thick particle in the left ventricle (LV) cavity behind the posterior leaflet and mild mitral regurgitation (MR). No aortic insufficiency (AI) or pulmonary vein (PV) stenosis were also observed. In addition, high-resolution computed tomography (HRCT) revealed left pleural effusion, scattered sub-segmental atelectasis in both lungs, and bilateral mild pneumothorax. Due to the possibility of bullet embolization, the patient underwent elective surgery to remove the foreign body from inside the heart. The surgical procedure was conducted via the median sternotomy approach. A partial thymectomy and pericardial opening were performed. The quantity of blood and clots evacuated from the pericardium Although dissection of the mediastinum and the great vessels was challenging, it was executed without harm. The entry point of the bullet was found in the anterosuperior part of the lower right pulmonary vein, which was covered by a clot. After the right atriotomy, the interventricular septum was split, and the bullet stuck in the posterior chordal lobe of the mitral valve was released and removed (Figure 3). Then the septum between the atrium and right atrium and the pulmonary vein are damaged and repaired, and after coronary perfusion is established and normal heart contractions return, cardiopulmonary bypass is stopped and the chest wall is repaired (Figure 4). Following the closure of the sternum, the patient was transferred to the intensive care unit and was deemed to be in stable condition. The patient underwent extubation within the initial 24 h following the surgical procedure. The individual achieved full recuperation and was released from medical care after 14 days. During a subsequent 6-month evaluation, the patient continued to exhibit a state of well-being, and there were no symptoms or signs of cardiothoracic infection or evidence of MR in echocardiography.

3 Discussion

Missile embolism is a rare subtype of foreign body embolization that can occur as a complication of gunshot injuries. In such cases, the missile enters the vessel or cardiac cavity but loses its kinetic energy and remains within the lumen instead of passing through. Embolization becomes a concern when there is no exit wound for the projectile, and it can be located distant from its expected trajectory. While most instances occur shortly after injury, delayed occurrences ranging from days to years have also been reported in the literature (8, 9). The incidence of this phenomenon has been reported to be 0.3% in the Vietnam War and 1.1% in more recent conflicts, such as the Afghanistan and Iraq Wars. However, its occurrence in civilian populations remains unknown, although it may be higher than in military settings due to the lower velocity and kinetic energy of civilian weapons (1, 10, 11). As previously mentioned, most venous emboli migrate with the direction of blood flow and are typically found in the pulmonary arterial system or right heart (12). Missile emboli to the left heart are infrequent and occur via

direct injury, entry through the pulmonary venous system, or a patent foramen oval (13). Symbas conducted a literature review spanning from 1940 to 1988 and found that out of the 201 patients with retained missiles in their hearts, the majority (87%) were due to direct injury to the heart or pericardium. The remaining 13% ($n=27$) were caused by an embolization originating from a distant site of injury. Of those, 81% had an injury occur within a systemic vein resulting in embolization to the right heart, while only one patient experienced damage to a pulmonary vein leading to embolization on the left side of the heart. For the remainder of the cases studied, it was unclear where exactly entry into circulation occurred (12). According to Advanced Trauma Life Support principles, stabilization is crucial in penetrative chest trauma. This involves performing life-saving interventions such as endotracheal intubation, placement of chest tubes, transfusion of blood products, or emergency department thoracotomy. In an unstable patient, even during the initial assessment, a chest x-ray study is vital because it provides information concerning the location and potential trajectory of a projectile if present, which can determine if there is the absence or presence of pneumothorax or hemothorax. The presence of a blurred appearance around the heart on an x-ray indicates a movement that may indicate intracardiac or pericardial positioning (14). Patients with stable hemodynamics presenting with projectiles within their hearts pose diagnostic and therapeutic challenges that require accurate localization and evaluation for associated injuries using multidisciplinary approaches, including surgical intervention radiographic analysis, endoscopy, or bronchoscopy. Using a CT scan represents an ideal imaging modality to localize projectiles accurately and ensure evaluation for associated injuries, including pulmonary parenchyma injury assessment and thoracic vascular structures assessment; whereas tracheobronchial tree damage on top esophagus injury judging is possible with this method (15, 16). In cases where there are apprehensions regarding injuries to the aforementioned structures, it is recommended to employ esophagoscopy or esophagogram and bronchoscopy to evaluate the esophagus and tracheobronchial tree, respectively. An intracardiac projectile can be localized through various modalities, such as TTE (transthoracic echocardiography), TEE, fluoroscopy, and coronary angiography. TTE is useful for unstable patients, as it can quickly confirm or rule out the presence of a pericardial effusion that may require operative intervention (17). However, TEE is superior to TTE in determining the precise location of the projectile and the extent of myocardial damage, injury to valves, or septum. Fluoroscopy may be helpful when echocardiography fails to determine bullet location accurately (18–20). The control of intracardiac bullets in stable patients is a topic of debate within the medical community due to the challenges in predicting long-term outcomes, which can vary from asymptomatic to severe and life-threatening. The occurrences related to a projectile are influenced by various factors such as the site, size, shape, and mobility. It is imperative to tailor treatment to the specific characteristics of the projectile (21) for instance, Nagy et al. expressed that tiny projectiles measuring 5 mm, situated bilaterally adjacent to the heart,

characterized by their smooth contours and complete intramyocardial embedding, typically exhibit no symptoms and may be conservatively managed without intervention (22).

4 Conclusion

The removal of intracardiac missiles is not always necessary. A limited number of patients who present at medical facilities with vital signs after experiencing penetrating trauma and bearing a foreign object within the heart necessitate expeditious resuscitation and hemodynamic stabilization. Management strategies should be tailored to each specific case. If there are missiles that are completely embedded in the heart and do not cause any symptoms, they can be monitored without intervention. On the other hand, any missiles that cause symptoms should be removed. Missiles that are asymptomatic and located on the left side of the heart should be removed to prevent systemic embolization and potential peripheral or cerebral ischemia. Likewise, asymptomatic missiles on the right side of the heart should be removed to prevent pulmonary emboli if they are larger than 5 mm. However, if a right-sided missile has already embolized the pulmonary circulation but is located peripherally and the patient remains asymptomatic, it can be observed without intervention (23).

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 requirement of ethical approval was waived by Iran University Medical of Sciences for the studies involving humans. 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

MK: Writing – review & editing. FK: Writing – original draft. MG: Writing – review & editing. HT: 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 our colleagues for their clinical management.

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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

References

- Rich NM, Collins GJ Jr, Andersen CA, McDonald PT, Kozloff L, Ricotta JJ. Missile emboli. *J Trauma Acute Care Surg.* (1978) 18(4):236–9. doi: 10.1097/00005373-197804000-00002
- Mattox KL, Beall AC Jr, Ennix CL, DeBakey ME. Intravascular migratory bullets. *Am J Surg.* (1979) 137(2):192–5. doi: 10.1016/0002-9610(79)90143-0
- Yoon B, Grasso S, Hofmann LJ. Management of bullet emboli to the heart and great vessels. *Mil Med.* (2018) 183(9–10):e307–13. doi: 10.1093/milmed/usx191
- Bartlett H, Anderson CB, Steinhoff NG. Bullet embolism to the heart. *J Trauma Acute Care Surg.* (1973) 13(5):476–9. doi: 10.1097/00005373-197305000-00011
- Fisk RL, Addetia A, Gelfand ET, Brooks CH, Dvorkin J. Missile migration from lung to heart with delayed systemic embolization. *Chest.* (1977) 72(4):534–5. doi: 10.1378/chest.72.4.534
- Braun SD. Arterial embolization of a bullet. *AJR Am J Roentgenol.* (2003) 180(1):281. doi: 10.2214/ajr.180.1.1800281
- Bland EF, Beebe GW. Missiles in the heart: a twenty-year follow-up report of world war II cases. *N Engl J Med.* (1966) 274(19):1039–46. doi: 10.1056/NEJM196605122741901
- Bakir DA, Othman YN, Taha AY. Pulmonary bullet embolism following cardiac gunshot wound: case report of a bullet that traversed the heart twice. *Cardiothorac Surg.* (2020) 28:1–5. doi: 10.1186/s43057-020-0017-5
- Michelassi F, Pietrabissa A, Ferrari M, Mosca F, Vargish T, Moosa H. Bullet emboli to the systemic and venous circulation. *J Br Surg.* (1990) 77(4):466–72. doi: 10.1002/bjs.1800770432
- Nolan T, Phan HO, Hardy AH, Khanna P, Dong P. Bullet embolization: multidisciplinary approach by interventional radiology and surgery. *Seminars in Interventional Radiology.* New York, NY: Thieme Medical Publishers (2012). Vol. 29, No. 3, p. 192–6. doi: 10.1055/s-0032-1326928
- Bartlett CS. Clinical update: gunshot wound ballistics. *Clin Orthop Relat Res.* (2003) 408:28–57. doi: 10.1097/00003086-200303000-00005

12. Symbas PN, Picone AL, Hatcher C, Vlasis-Hale S. Cardiac missiles. A review of the literature and personal experience. *Ann Surg.* (1990) 211(5):639. PMCID: PMC1358242
13. Schurr M, McCord S, Croce M. Paradoxical bullet embolism: case report and literature review. *J Trauma Acute Care Surg.* (1996) 40(6):1034–6. doi: 10.1097/00005373-199606000-00034
14. Nguyen V, Nguyen K. Plain film of intracardiac foreign bodies: the blurring effect. *South Med J.* (1991) 84(5):651–3. PMID: 2035092
15. Gandhi SK, Marts BC, Mistry BM, Brown JW, Durham RM, Mazuski JE. Selective management of embolized intracardiac missiles. *Ann Thorac Surg.* (1996) 62(1):290–2. doi: 10.1016/0003-4975(96)00097-5
16. Nagy KK, Roberts RR, Smith RF, Joseph KT, An GC, Bokhari F, et al. Trans-mediastinal gunshot wounds: are “stable” patients really stable? *World J Surg.* (2002) 26(10):1247–50. doi: 10.1007/s00268-002-6522-2
17. Freshman SP, Wisner DH, Weber CJ. 2-D echocardiography: emergent use in the evaluation of penetrating precordial trauma. *J Trauma Acute Care Surg.* (1991) 31(7):902–6. doi: 10.1097/00005373-199107000-00004
18. Hashimi MW, Jenkins DR, McGwier BW, Massey CV, Alpert MA. Comparative efficacy of transthoracic and transesophageal echocardiography in detection of an intracardiac bullet fragment. *Chest.* (1994) 106(1):299–300. doi: 10.1378/cheest.106.1.299
19. Limandri G, Gorenstein LA, Starr JP, Homma S, Auteri J, Gopal AS. Use of transesophageal echocardiography in the detection and consequences of an intracardiac bullet. *Am J Emerg Med.* (1994) 12(1):105–6. doi: 10.1016/0735-6757(94)90214-3
20. Fragomeni L, Azambuja PC. Bullets retained within the heart: diagnosis and management in three cases. *Thorax.* (1987) 42(12):980–3. doi: 10.1136/thx.42.12.980
21. Symbas PN, Symbas PJ. Missiles in the cardiovascular system. *Chest Surg Clin N Am.* (1997) 7(2):343–56. PMID: 915629
22. Nagy KK, Massad M, Fildes J, Reyes H. Missile embolization revisited: a rationale for selective management. *Am Surg.* (1994) 60(12):975–9. PMID: 7992978
23. Galante J, London JA. Left ventricular bullet embolus: a case report and review of the literature. *J Emerg Med.* (2010) 39(1):25–31. doi: 10.1016/j.jemermed.2007.09.057



OPEN ACCESS

EDITED BY

Giuseppe Gatti,
Azienda Sanitaria Universitaria Giuliano
Isontina, Italy

REVIEWED BY

Jayant Shyam Jainandunsing,
University Medical Center Groningen,
Netherlands
Tommaso Hinna Danesi,
University of Cincinnati Medical Center,
United States

*CORRESPONDENCE

Huanlei Huang
✉ hhuanlei@hotmail.com

¹These authors have contributed equally to
this work

RECEIVED 23 September 2023

ACCEPTED 06 February 2024

PUBLISHED 26 February 2024

CITATION

Wang Z, Zhong L, Guo H, Liu Y, Zhou C, Ye Y, Han F and Huang H (2024) Case Report:
Totally endoscopic minimally invasive mitral
valve surgery during pregnancy: a case series.
Front. Cardiovasc. Med. 11:1300508.
doi: 10.3389/fcvm.2024.1300508

COPYRIGHT

© 2024 Wang, Zhong, Guo, Liu, Zhou, Ye, Han
and Huang. This is an open-access article
distributed under the terms of the [Creative
Commons Attribution License \(CC BY\)](#). The
use, distribution or reproduction in other
forums is permitted, provided the original
author(s) and the copyright owner(s) are
credited and that the original publication in
this journal is cited, in accordance with
accepted academic practice. No use,
distribution or reproduction is permitted
which does not comply with these terms.

Case Report: Totally endoscopic minimally invasive mitral valve surgery during pregnancy: a case series

Zhenzhong Wang^{1†}, Lishan Zhong^{1†}, Haijiang Guo¹, Yanli Liu²,
Chengbin Zhou¹, Yingxian Ye³, Fengzhen Han² and
Huanlei Huang^{1*}

¹Department of Cardiac Surgery, Guangdong Provincial People's Hospital (Guangdong Academy of Medical Sciences), Southern Medical University, Guangzhou, China, ²Department of Obstetrics, Guangdong Provincial People's Hospital (Guangdong Academy of Medical Sciences), Southern Medical University, Guangzhou, China, ³Department of Anesthesia, Guangdong Provincial People's Hospital (Guangdong Academy of Medical Sciences), Southern Medical University, Guangzhou, China

A totally endoscopic minimally invasive approach is widely used for cardiac valve surgery in normal adults. However, minimally invasive cardiac surgery during pregnancy is rarely reported. In addition to traditional median thoracotomy, totally endoscopic minimally invasive approaches can now be used for pregnant patients. We describe our experience with totally endoscopic cardiac valve surgery (TECVS) during pregnancy, which is safe for both mothers and fetuses.

KEYWORDS

total endoscopy, cardiac valve surgery, pregnancy, cardiopulmonary bypass, minimally invasive

Introduction

Heart disease is the primary cause of maternal and fetal death in 1%–4% of pregnancies (1). Cardiac surgery is recommended only when medical therapy or interventional procedures fail and the mother's life is threatened (2). This is likely due to the difficulty of performing cardiac surgery during pregnancy and the complexity of multidisciplinary cooperation with important departments, such as obstetrics. We herein introduce our approach to TECVS during pregnancy.

Description of the cases

In all patients, general anesthesia was managed. Patients were placed in the 20° counterclockwise lateral decubitus position with a mat under the right shoulder. The right upper arm was positioned horizontally to the thorax. A venous cannula was placed in the superior cava vein through the jugular vein by an anesthesiologist. The cannula for cardiopulmonary bypass (CPB) was passed through the femoral vessel. After a 2 cm–2.5 cm long skin incision was made in the right groin, the femoral vein was sutured to the femoral artery with one layer of 3-0 Prolene sutures and two layers of purse sutures before puncture at the midpoint under transesophageal echocardiography (TOE) guidance. The guidewires were removed after TOE-guided

confirmation that the venous catheter had been placed in the right atrium and that the arterial catheter had been placed in the ascending aorta.

The 3-port minimally invasive cardiac surgery technique was mastered (Figure 1A). The system includes a main working port, an auxiliary working port and one camera port. This technique is significantly less likely to damage the mammary gland. A two-dimensional (30° oblique view) thoracoscope was inserted through the camera port. The aortic cannula was placed through the main working port, and the left heart drain was placed through the auxiliary working port (Figure 1B). Fetal heart rate was monitored throughout the procedure (Figures 1C,D). The technique allows real-time intraoperative monitoring of specific fetal heart rate changes as well as contraction pressures.

To reduce the impact of CPB, we did not start CPB until the aortic purse string suture was completed. We proposed pulsatile flow perfusion, normothermic perfusion ($>35^{\circ}\text{C}$) and high-flow rate ($>2.5 \text{ L/min/m}^2$) perfusion. In addition, phenylephrine can be used to maintain a systolic pressure $\geq 70 \text{ mmHg}$ to ensure placental perfusion. To minimize the duration of the procedure as much as possible, experienced surgeons and nurses must collaborate effectively during surgery. Patients were weaned from CPB as soon as possible, and the femoral vessel cannula was removed. Warfarin was used for anticoagulation, and the international normalized ratio (INR) was monitored weekly or every 2 weeks. Low-molecular-weight heparin was used for one week before delivery.

Results

From January 2019 to September 2022, a total 9 patients, including 4 advanced age mothers, underwent totally endoscopic cardiac valve surgery (Table 1). Eight patients were categorized as NYHA class II, and one patient was NYHA class III. Mitral valve replacement was performed in 6 (66.7%) patients. One (11.1%) patient underwent mitral valve repair, tricuspid repair and left atrial appendage closure (Figures 2A–C). The CPB time, aortic cross-clamp (ACC) time and thoracic drainage volume during the first 24 h after surgery were $89.7 \pm 13.4 \text{ min}$, $58.3 \pm 11.5 \text{ min}$ and $260.0 \pm 126.1 \text{ ml}$, respectively. The durations of mechanical ventilation, intensive care unit (ICU) stay and postoperative hospital stay were $3.7 \pm 2.9 \text{ h}$, $33.3 \pm 22.8 \text{ h}$ and $5.3 \pm 2.2 \text{ days}$, respectively. Six (66.7%) pregnant women delivered via cesarean section. Only one of the newborns was born prematurely (34^{+5} weeks). Both the 1- and 5-minute APGAR scores for all the newborns was 10. Vaginal bleeding occurred in the operating room immediately after surgery in a pregnant woman (16^{+3} weeks), and fetal ultrasound revealed no fetal heart rate. In addition, there were two cases of fetal loss after discharge. Two months after surgery, labor was induced for Patient No. 1 due to fetal nervous system abnormalities. Patient no. 2 underwent induction of labor because of personal reasons 6 days after surgery (Table 2).

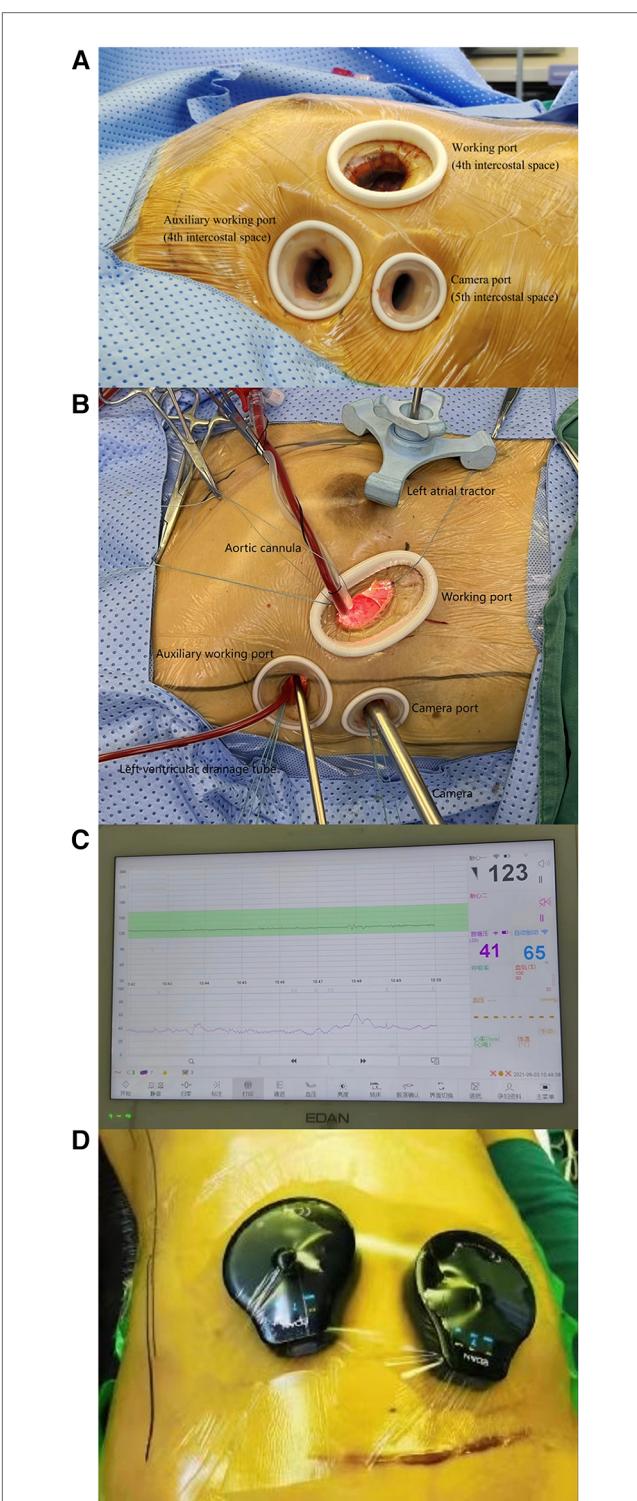


FIGURE 1

The figure shows the spatial layout of the 3-port minimally invasive cardiac surgery technique (A,B). Continuous fetal heart rate monitoring during surgery (C,D).

Discussion

Compared with median thoracotomy, TEVAR has more advantages for normal patients, such as smaller wounds, less

TABLE 1 Preoperative details of the individual patients.

Patient no.	NYHA class	mWHO class	Age (years)	GA (weeks)	Cause of disease	Diagnosis	LVEF (%)	PAP (mmHg)
1	II	IV	28	21 ⁺⁶	IE	Severe MR, moderate MS	60	80
2	II	IV	39	18 ⁺⁶	RHVD	Severe MS, moderate MR	62	45
3	II	IV	37	18 ⁺⁵	RHVD	Moderate to severe MS, Moderate to severe MR	63	62
4	III	IV	34	32	RHVD	Severe MS, mild MR	76	60
5	II	IV	28	24	RHVD	Moderate to severe MR, moderate MS	72	57
6	II	IV	43	23	RHVD	Severe MR, mild MS	73	54
7	II	IV	36	21	RHVD	Moderate MS, moderate MR, severe TR, PFO	59	37
8	II	IV	28	16 ⁺²	RHVD	Extremely severe MR	65	48
9	II	II-III	33	27 ⁺⁴	RHVD	Severe MR	74	30

NYHA, New York Heart Association; GA, gestational age; IE, infective endocarditis; RHVD, rheumatoid heart valve disease; MR, mitral regurgitation; MS, mitral stenosis; TR, tricuspid regurgitation; PFO, patent foramen ovale; PAP, pulmonary arterial pressure.

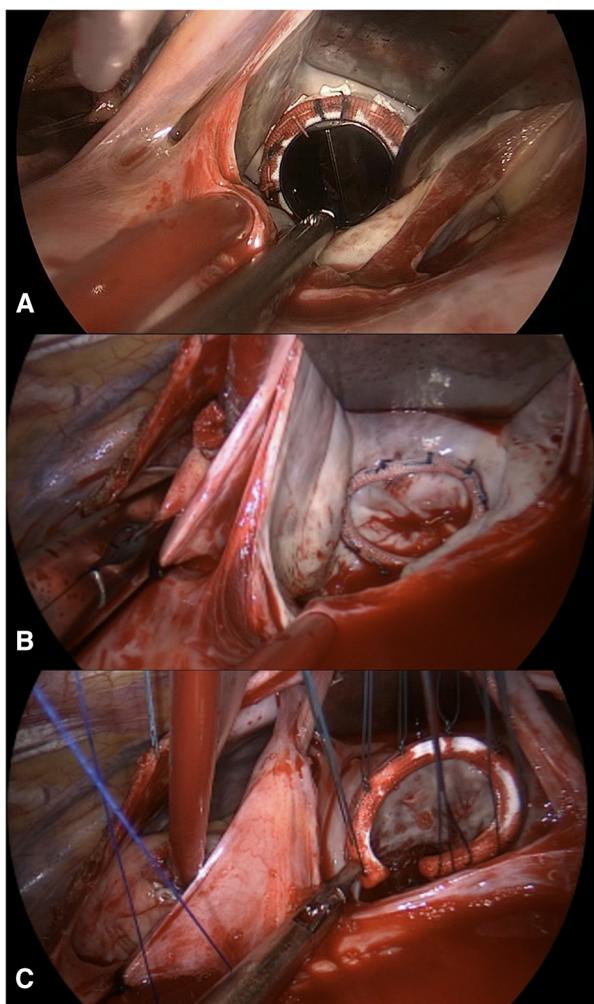


FIGURE 2
The figure shows the results of mitral valve replacement (A), mitral annuloplasty (B), and tricuspid annuloplasty (C).

pain, less blood loss and shorter hospital stays (3). TECVS is a suitable option for pregnant women. The integral sternum allows better upper extremity movement and does not restrict weight

lifting so that the mother can feed the baby (4). Moreover, scars were less noticeable. In addition, the efficacy of TECVS was not affected by the patient's weight, especially obese pregnant patients, and the mitral and tricuspid valves are better exposed in TECVS (5). Moreover, less postoperative pain is good for maintaining intrauterine pregnancy, which is critical for fetal survival and growth.

Pregnant patients and fetuses are significantly less tolerant of cardiac surgery than healthy adults are. The timing of surgery is crucial. In terms of fetal safety, if the operation starts too early, fetal teratogenesis (6) can easily occur, possibly leading to premature birth. The best period for surgery is between 13 and 28 weeks of gestation (2). However, it is worth noting that we successfully performed the surgery 32 weeks of gestation, and a full-term fetus was successfully delivered. However, one patient underwent surgery at 16 weeks of gestation, at which point the fetus died. Therefore, the best period for surgery deserves further study.

According to the modified World Health Organization (WHO) classification, all patients were class IV, which indicated that the maternal cardiac event rate was 40%–100% (2). The fetal mortality rate during CPB remains high (~20%) (7). There are three unique pathophysiological changes in pregnancy during CPB: uterine contraction, placental hypoperfusion and fetal hypoxia (8). Most researchers agree that pulsatile flow perfusion, normothermic perfusion (>35°C) and high-flow rate (>2.5 L/min/m²) perfusion are beneficial². Blood pressure was unstable according to our observations. We propose using phenylephrine to maintain a systolic pressure ≥70 mmHg to ensure placental perfusion. With the cooperation of perfusionists, it was feasible to start CPB just before ACC and end CPB as soon as possible. This approach was the most direct way to reduce CPB time.

The primary aim of TECVS is to cure cardiac valve disease while also protecting the fetus. The fetal mortality rate for cardiac surgery in pregnancy is approximately 30% (9). In our case series, 1 fetus (No. 8) died during surgery. We believe that this may be because both her CPB time and ACC time were above average and because she was in the early weeks of pregnancy. The fetuses of the other two patients died during the follow-up period. Patient No. 1 had fetal nervous system

TABLE 2 Results of individual patients.

Patient No.	Surgical procedure	CPB (min)	ACC (min)	MV (h)	ICU (h)	PODS (d)	Outcome	
							Maternal	Fetal
1	MVR (STJ.#25)	92	64	5	39	5	Survived	Died, 35 weeks
2	MVR (STJ.#27)	75	47	4	22	8	Survived	Died, 20 weeks
3	MVP (EL.#26)	90	69	6	22	5	Survived	Alive, full term
4	MVR (Lanfei.#27)	67	43	10	22.5	3	Survived	Alive, full term
5	MVP (EL.#26)	77	45	3	29	5	Survived	Alive, full term
6	MVR (EL.#27)	96	57	0	37	4	Survived	Alive, premature
7	MVP (EL.#30)	96	57	1	19	5	Survived	Alive, full term
	TVP (EL.#28)							
	LAAO, PFOC							
8	MVP	102	63	40	94	10	Survived	Died, the day after surgery
9	MVP	112	80	2	15	3	Survived	Alive, full term

CPB, cardiopulmonary bypass; ACC, aortic cross-clamp; MV, mechanical ventilation; ICU, intensive care unit; PODS, postoperative days; PFOC, patent foramen ovale closure; MVR, mitral valve replacement; MVP, mitral valvuloplasty; TVP, tricuspid valvuloplasty; LAAO, left atrial appendage occlusion.

abnormalities. This case is the first in her and her husband's immediate family. She had been taking low-dose warfarin during her pregnancy. We believe that this patient's warfarin use may have caused fetal death, but we lack additional imaging evidence. Fetal death and fetal warfarin syndrome have also been reported with low-dose warfarin (10). The death of fetus No. 2 was chosen by the patient of her own accord, and there was no abnormality in the fetus at the time.

A multidisciplinary team is best suited for providing optimal support during the procedure. The participants included cardiac surgeons, obstetricians, anesthesiologists, perfusionists and nurse specialists. Anesthesiologists were responsible for ensuring a NRS score <3. Obstetricians immediately assessed the condition of the fetus after surgery and examined the patient in the hospital at least once per day. Atosiban acetate was injected to suppress uterine contractions when necessary. After discharge, transthoracic echocardiography was recommended for pregnant patients. All patients gave birth at our medical center under the care of a multidisciplinary team.

Conclusions

To our knowledge, cardiac valve surgery during pregnancy poses a serious challenge. Our report suggested that TECVS can be an option for pregnant patients and that fetuses could be maintained with intrauterine pregnancy. Studies with more patients and longer follow-up periods will be performed in the future.

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 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

ZW: Writing – original draft. LZ: Data curation, Writing – review & editing. HG: Methodology, Writing – review & editing. YL: Investigation, Writing – review & editing. CZ: Writing – review & editing. YY: Writing – review & editing. FH: Data curation, Writing – review & editing. HH: Methodology, Writing – review & editing, Funding acquisition, Writing – original draft.

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. 82270373).

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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated

organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

References

1. Liu Y, Han F, Zhuang J, Liu X, Chen J, Huang H, et al. Cardiac operation under cardiopulmonary bypass during pregnancy. *J Cardiothorac Surg.* (2020) 15:92. doi: 10.1186/s13019-020-01136-9
2. Regitz-Zagrosek V, Roos-Hesselink JW, Bauersachs J, Blomström-Lundqvist C, Cifková R, De Bonis M, et al. 2018 ESC guidelines for the management of cardiovascular diseases during pregnancy. *Eur Heart J.* (2018) 39:3165–241. doi: 10.1093/eurheartj/ehy340
3. Chen Y, Huang L-C, Chen D-Z, Chen L-W, Zheng Z-H, Dai X-F. Totally endoscopic mitral valve surgery: early experience in 188 patients. *J Cardiothorac Surg.* (2021) 16:91. doi: 10.1186/s13019-021-01464-4
4. Taksaudom N, Traisrisilp K, Kanjanavanit R. Left atrial myxoma in pregnancy: management strategy using minimally invasive surgical approach. *Case Rep Cardiol.* (2017) 2017:8510160. doi: 10.1155/2017/8510160
5. Nguyen S, Umana-Pizano JB, Donepudi R, Dhoble A, Nguyen TC. Minimally invasive mitral valve repair for acute papillary muscle rupture during pregnancy. *Ann Thorac Surg.* (2019) 107:e93–95. doi: 10.1016/j.athoracsur.2018.06.048
6. Parry AJ, Westaby S. Cardiopulmonary bypass during pregnancy. *Ann Thorac Surg.* (1996) 61:1865–9. doi: 10.1016/0003-4975(96)00150-6
7. Kapoor MC. Cardiopulmonary bypass in pregnancy. *Ann Card Anaesth.* (2014) 17:33–9. doi: 10.4103/0971-9784.124133
8. Yuan S-M. Indications for cardiopulmonary bypass during pregnancy and impact on fetal outcomes. *Geburtshilfe Frauenheilkd.* (2014) 74:55–62. doi: 10.1055/s-0033-1350997
9. Shook LL, Barth WH. Cardiac surgery during pregnancy. *Clin Obstet Gynecol.* (2020) 63:429–46. doi: 10.1097/GRF.0000000000000533
10. Basu S, Aggarwal P, Kakani N, Kumar A. Low-dose maternal warfarin intake resulting in fetal warfarin syndrome: in search for a safe anticoagulant regimen during pregnancy. *Birth Defects Res A Clin Mol Teratol.* (2016) 106:142–7. doi: 10.1002/bdra.23435



OPEN ACCESS

EDITED BY

Giuseppe Gatti,
Azienda Sanitaria Universitaria Giuliano
Isontina, Italy

REVIEWED BY

Weichieh Lee,
Chi Mei Medical Center, Taiwan
José María Arribas,
Virgen de la Arrixaca University Hospital, Spain

*CORRESPONDENCE

Mohammadbagher Sharifkazemi
✉ dr.sharifkazemi@gmail.com

RECEIVED 24 January 2024

ACCEPTED 02 May 2024

PUBLISHED 20 May 2024

CITATION

Sharifkazemi M, Ghazinour M, Lotfi M, Khorshidi S and Davarpasand T (2024) Retrosternal hematoma causing torsade de pointes after coronary artery bypass graft surgery; a case report. *Front. Cardiovasc. Med.* 11:1331873. doi: 10.3389/fcvm.2024.1331873

COPYRIGHT

© 2024 Sharifkazemi, Ghazinour, Lotfi, Khorshidi and Davarpasand. This is an open-access article distributed under the terms of the [Creative Commons Attribution License \(CC BY\)](#). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

Retrosternal hematoma causing torsade de pointes after coronary artery bypass graft surgery; a case report

Mohammadbagher Sharifkazemi^{1*} , Mohammad Ghazinour², Mehrzad Lotfi³, Soorena Khorshidi¹ and Tahereh Davarpasand⁴

¹Department of Cardiology, Nemazee Hospital, Shiraz University of Medical Sciences, Shiraz, Iran,

²Department of Surgery, Shahid Faghihi Hospital, Shiraz University of Medical Sciences, Shiraz, Iran,

³Department of Radiology, Shahid Faghihi Hospital, Shiraz University of Medical Sciences, Shiraz, Iran,

⁴Department of Cardiology, Tehran Heart Center, Tehran University of Medical Sciences, Tehran, Iran

Myocardial infarction is among the top causes of mortality worldwide. Survivors may also experience several complications. Infarct-related torsade de pointes (TdP) is an uncommon complication. In the context of myocardial infarction, coronary artery bypass graft (CABG) surgery is the prevalent therapeutic modality associated with several early and late complications. Ventricular tachyarrhythmias, including TdP, because of electrical inhomogeneity, would potentially be a lethal complication of CABG. Here, we report the occurrence of medically intractable TdP in the presence of an uncommon case of a post-CABG retrosternal hematoma. Arrhythmia was properly resolved after hematoma removal surgically. It showed the possibility of a "cause and effect" relationship between these two complications. This unique case emphasizes the post-CABG medically-resistant TdP, considering the mechanical pressure effect of retrosternal hematoma that stimulates this potentially malignant arrhythmia, especially in the absence of electrolyte disturbances and evident symptoms of ongoing ischemia.

KEYWORDS

myocardial infarction, coronary artery bypass, torsades de pointes, cardiac tamponade, case report

Introduction

Myocardial infarction (MI), especially ST-elevation MI (STEMI), is among the top causes of mortality worldwide, responsible for at least 15% of mortality each year (1). Although emergency management, especially coronary artery bypass grafting (CABG) and percutaneous intervention, reduced its mortality rate (2), several complications are observed in the survivors, such as post-MI mechanical complications and electrical disturbances (3). Complications are sorted into early and late events, most requiring emergency interventions (4).

Arrhythmias and conduction disturbances, such as ventricular fibrillation (VF), ventricular tachycardia (VT), and many other arrhythmias, are not uncommon complications, both in early and late stages even after CABG that needs medical and sometimes surgical intervention (5). Polymorphic VT in the setting of acute MI is commonly observed; however, acquired long QT syndrome infarct-related torsade de pointes (TdP) is uncommon (6). Because this complication is potentially lethal, paying greater attention to its manifestations and associated clinical signs and symptoms is

important for accurate and on-time diagnosis and treatment. Considering the few cases reported in the literature, reporting characteristics and outcomes of new cases can help increase the physician's knowledge about this lethal complication in the post-CABG status. Accordingly, in the present study, we report a drug-resistant TdP in the presence of retrosternal hematoma as an early complication after CABG, which frequently stimulated arrhythmias, and finally stopped after hematoma removal surgically.

Case presentation

The patient was a 69-year-old man with a positive history of diabetes mellitus, admitted to the emergency department with the chief complaint of retrosternal pain, accompanied by nausea and dyspnea, for several hours before admission. He was diagnosed with an inferolateral ST-elevation MI, accompanied by a prolonged QTc interval on the first electrocardiogram (ECG) (Figure 1) and transferred to the catheterization laboratory for emergency coronary angiography, which revealed significant stenosis in the terminal end of the left main artery, in addition to advanced three-vessel disease. Accordingly, CABG was recommended.

He was admitted to the cardiac surgery ward for about 10 days to control his blood sugar and become prepared for the surgery. During this period, no ventricular arrhythmia was recorded. The serum levels of potassium, magnesium, and calcium were evaluated, all of which were in the normal range. The follow-up echocardiography showed dilated left ventricle (LV) with global hypokinesia and severe LV dysfunction, LV ejection fraction (EF) of 25%, a huge apical aneurysm, a large semi-mobile apical clot (its area was equal to 2.4 cm^2), and grade II diastolic dysfunction.

Accordingly, CABG (one arterial and two vein graft LIMA-LAD, SVG-OM, and SVG-PDA) was done with LV aneurysmectomy, LV clot removal, and intra-aortic balloon pump (IABP) insertion. Aortic clamp time was 50 min, cardiopulmonary bypass (CPB) time was 90 min, and total operation duration was 200 min. Two chest tubes were inserted for the patient in the substernal and left pleural spaces. After the surgery, he was transferred to the cardiac surgery intensive care unit (ICU). The post-CABG ECG of the patient (in ICU) illustrated normal sinus rhythm with low voltage QRS, and bigeminal ventricular extrasystole (PVCs) with short coupling interval (R on T wave; Supplementary Figure S1).

The day after the surgery, the serum level of hemoglobin was 9 mg/dl, and he was transfused with one unit (400 ml) packed cell. On the same day, anti-platelet therapy started, and IABP was removed (24 h after the surgery). On the first day 600 ml was darianed, and on next day (48 h after the surgery), 100 ml; the chest tube was removed after 48 h. On the same day (2 days after the surgery), he developed recurrent episodes of TdP (Figure 2), one of them degenerated into VF, causing cardiovascular collapse. Hence, he was re-intubated and received 200 J DC shock, in addition to 2 g IV lidocaine plus 24 g IV magnesium sulfate in a 24-h infusion. Moreover, because of persistent low systolic blood pressure, IABP was re-inserted. The serum levels of cardiac enzymes (such as high sensitivity troponin I) and the electrolytes were normal. The patient's LVEF was 25%–30% on the third postoperative day.

After a couple of days, IABP was removed, and the patient was extubated. While serum levels of electrolytes were within the normal range, long QTc remained. Accordingly, IV infusion of magnesium sulfate and lidocaine continued for more following 5

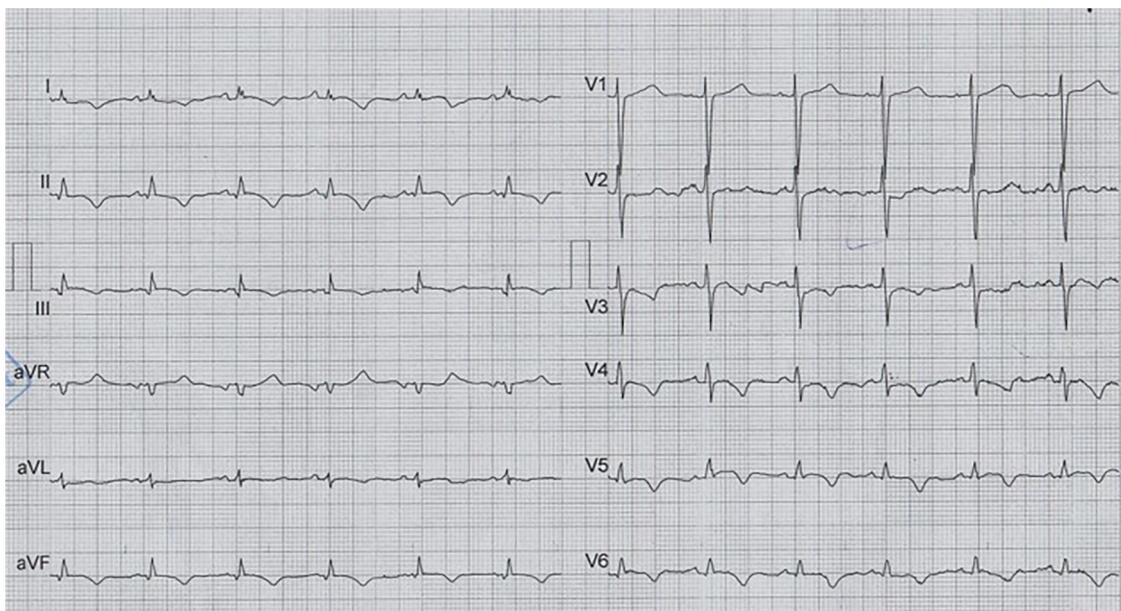


FIGURE 1

The first ECG on arrival and before CABG, which illustrates normal sinus rhythm with low voltage QRS and prolonged QTcB (505 ms), in addition to findings compatible with inferolateral STEMI.

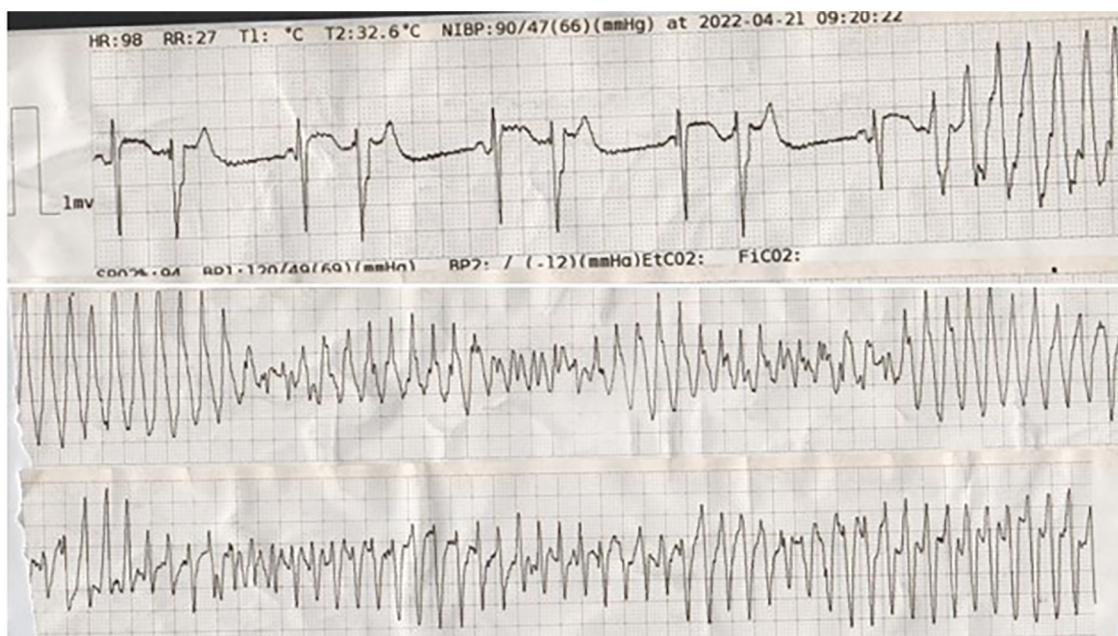


FIGURE 2

The tracing shows bigeminal PVCs with short coupling intervals (R on T), which degenerated into torsade de pointes.

days. Several episodes of sustained and non-sustained TdPs were noted during this period, managed by DC shocks when sustained, resulting in decreased blood pressure. These treatment modalities helped the patient return to sinus rhythm each time. Meanwhile, a bedside transthoracic echocardiography by an expert cardiologist echocardiographer showed a large-size retrosternal hematoma, which extended to the apicolateral of the RV with a compression effect over the RV-free wall. A chest computed tomography (CT) with intravenous contrast was

performed for a more accurate evaluation of the hematoma. Post-contrast scan illustrated a high soft tissue density lesion in the anterior mediastinum with Hounsfield unit (HU) consistent with clotted blood (Figure 3). After retrosternal clot removal by the second surgery, the patient's malignant arrhythmia stopped without any other interventions. Antiarrhythmic drugs stopped and the patient stayed in the hospital for another 2 weeks for close follow-up. Considering in-hospital repeated episodes of TdP before the second surgery, as well as persistent long QTc post-

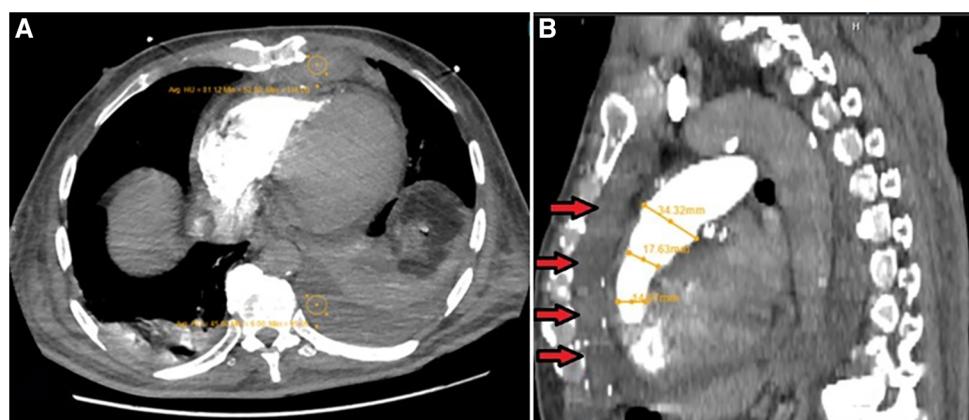


FIGURE 3

(A) Post-contrast axial CT scan during pulmonary arterial phase, which shows high soft tissue density lesion in the anterior mediastinum with hounsfield unit (HU) consistent with clotted blood (HU = 81). Also note the presence of mild left-sided bloody pleural effusion (HU = 45), associated with the underlying collapse of the posterior basal segment of the left lower lobe and a lesser degree of segmental atelectasis of the posterior basal segment of the right lower lobe, all secondary to recent CABG operation. (B) MPR images show narrowing of RVOT secondary to external pressure effect by retrosternal hematoma (arrows).

second surgery, he underwent uneventful single chamber implantable cardioverter defibrillator (ICD) implantation. During his 12-month follow-up, ICD analysis showed no high ventricular rate, despite the existence of long QT on several ECGs taken during this follow-up period. Historical care is organized as a timeline (Table 1).

Considering the patient's perspective, he appeared cooperative and established a friendly patient-physician relationship. He had some extent of anxiety, considering the persistent arrhythmia that resulted in longer hospitalization; but after the second surgery that resulted in clinical improvement, the patient's anxiety faded away, and he went home satisfied with the care he received.

Discussion

Here, we reported a patient with TdP arrhythmia after CABG, resolved by the removal of the retrosternal hematoma. Considering the wide range of factors inducing TdP after CABG, we reported this case to emphasize that the etiology of arrhythmia has to be discovered first, and the treatment should be selected accordingly. Supraventricular arrhythmias, especially atrial fibrillation, are very common after cardiac surgery and affect the prognosis and duration of hospital stay (7, 8). Similarly, ventricular arrhythmias are also observed secondary to MI and/or CABG with several

patient- and surgery-related risk factors (9). Finding the cause of arrhythmia formation can help the physician to treat it before it leads to fatal complications.

The type of arrhythmia in the patient reported here was TdP, a rare malignant arrhythmia that is a specific form of polymorphic VT occurring in the context of QT prolongation (10). Interestingly, our patient presented with long QTc at the time of referral (before the surgery). A review of all cases (available in the literature) suggested cardiac surgery and craniotomies accounting for 40% of all cases with perioperative TdP (11). Reports suggest that TdP is probably not the direct effect of CABG, but related to CABG complications (12–14), such as ischemia (due to graft failure) (12) or co-administration of QT-prolonging drugs (like amiodarone) in the perioperative period (13, 15). It is important to remember that prolonged QTc is multifactorial and can occur by genetic and inherited causes, as well (14). But, the exact cause of pre-surgical QT prolongation was not clarified in our patient, because the genetic testing was not done (due to the patient's financial inability); also, the patient did not take any QT-prolonging drugs. QT-prolonging drugs are found responsible for at least one-third of cases with perioperative TdP (11); some patients who develop drug-induced TdP are silent carriers of gene mutations related to prolonged QTc (16). While TdP development as a result of medications (like terfenadine and cisapride) is a cause of withdrawal, many other

TABLE 1 Timeline of the historical care organized by time.

Date	Day	Clinical signs and symptoms	Imaging or intervention	Results	Physician's decision
24.01.2022	1	Retrosternal pain, nausea, dyspnea	ECG	ST-elevation MI + prolonged QT interval	Advised to undergo emergency coronary angiography
24.01.2022	1	The patient was admitted	Emergent coronary angiography	Stenosis in the terminal end of left main artery + advanced 3VD	CABG was recommended
24.01.2022–03.02.2022	1–11	Admitted to cardiac surgery ward for controlling blood sugar	Serum analysis	Normal serum level of potassium, magnesium, and calcium	Close observation in the ward
24.01.2022–03.02.2022	1–11	Blood sugar was controlled	Echocardiography	Dilated LV + hypokinesia and severe LV dysfunction, EF = 25%, apical aneurysm + apical clot, grade II diastolic dysfunction	CABG with LV aneurysmectomy
03.02.2022	11	Transferred to operation room	Surgery	CABG + aneurysmectomy + clot removal with IABP insertion	Transferred to intensive care unit
05.02.2022–10.02.2022	13–18	Recurrent episodes of TdP, resulting in VF and cardiovascular collapse	ECG	200 J DC shock + 2 gr IV lidocaine + 24 gr magnesium sulfate in 24-h infusion IABP was re-inserted	Follow-up
07.02.2022	15	Hemodynamically stabilized	IABP was removed, extubated	Normal serum level of electrolytes + remained long QT interval	Continue magnesium sulfate and lidocaine infusion for 5 days
05.02.2022–10.02.2022	13–18	Sustained and non-sustained TdP	DC shock + treatment of hypotension	Return to sinus rhythm each time, but relapsed	Echocardiography
10.02.2022	18	Sustained and non-sustained TdP	Echocardiography	Large retrosternal hematoma compressing the free wall of right ventricle	Chest CT
10.02.2022	18	Sustained and non-sustained TdP	Chest CT with contrast	high soft tissue density lesion in the anterior mediastinum with Hounsfield unit consistent with clotted blood	Second surgery
11.02.2022	19	Admitted to the operation room	Second surgery	Removal of retrosternal clot	Stay in hospital for 2 weeks
23.02.2022	31	Underwent ICD implantation		Uneventful ICD-VR implantation	OPD follow up
25.02.2022	33	Discharged from hospital		Plan ICD analysis and programming	Follow-up for a year
26.02.2023	A year later	OPD visit and ICD analysis every three months over the following year		No high ventricular rate had recorded over the one-year ICD follow up analysis	Follow-up

ECG, electrocardiography; MI, myocardial infarction; 3VD, three-vessel disease; LV, left ventricle; CABG, coronary artery bypass surgery; EF, ejection fraction; IABP, intra-aortic balloon pump; TdP, torsad de point; VF, ventricular fibrillation; CT, computed tomography.

medications (like amiodarone and ranolazine) are still on the market, as they mainly cause prolonged QTc, but rarely TdP. Bearing in mind that the cumulative effect of two QTc-prolonging agents increases the risk in the patient, it is necessary to consider drug interactions (mediated through cytochrome P540) in the perioperative period (17, 18). Other risk factors of prolonged QTc include female sex, higher age, electrolyte abnormalities, anorexia nervosa, heart conditions (such as bradycardia, left ventricular dysfunction, heart failure, mitral valve prolapse, and MI), and other medical conditions (like renal/hepatic dysfunction, hypokalemia, hypoglycemia, hypertension, diabetes mellitus, hypothyroidism, pituitary insufficiency, injury to the central nervous system, malnutrition, and obesity) (18–20). Uncommon causes, like coronary atherosclerotic plaque rupture after thoracic trauma, have also been reported (21). There are also reports of patients with subdural hematoma (following head trauma), found to have TdP during admission (22–24). Some have also reported TdP as the presenting sign of cerebral hemorrhage (25). Therefore, it is necessary to pay attention to the multiple risk factors of TdP development and prevent co-administration of factors that increase the risk of TdP development and consider cases who are more susceptible to this arrhythmia, in order to diagnose it at an early stage and implement therapeutic strategies before fatal complications occur.

The case presented here had none of the above-mentioned causes; but, another rare postoperative complication was observed that we speculate it as the main cause of TdP formation. In this case, imaging investigations (accurate examinations by echocardiography and CT scan) showed a post-CABG retrosternal hematoma, causing localized tamponade. Hematoma, caused by the leaking of blood from an aneurysmectomy/aneurysmorrhaphy site, is listed among common postoperative complications of CABG; but not as a cause of post-CABG TdP. Hematoma, alone, is an important complication, as it can rarely compress the heart and arteries and may even cause superior vena cava syndrome (26) and tamponade, which lead to shock and death in the patients (27). Therefore, diagnosis is essential; previous studies have suggested CT scan as an accurate diagnostic tool for post-CABG epicardial and retrosternal hematoma formation (28, 29). We could also detect the details of the developed hematoma by CT scan successfully. This case shows the necessity to closely observe any patient who develops hypotension after CABG. Fortunately, our case was diagnosed appropriately and saved by appropriate treatment.

The need for re-operation has been reported previously in patients with post-CABG hematoma (29–34). We also removed the hematoma with a second surgery. A notable issue in our patient was the resolution of TdP after this (second) surgery, which suggests that the development of TdP after the first surgery could have been caused by the pressure effect of hematoma on the heart or small coronary arteries, which could lead to ischemia. We did not observe a similar case reporting an association between retrosternal hematoma and medically intractable TdP. This finding [resolution of arrhythmia after the second surgery (hematoma removal)] in our case suggests a “cause and effect” relationship between hematoma and drug-resistant TdP, which is of great significance, considering the

challenging treatment of TdP (35). Therefore, post-CABG hematoma should also be considered in cases who have sustained or recurrent TdP arrhythmia.

Other rare points documented in the present case report include the time and site of the hematoma. Development of hematoma at the early postoperative phase has been only reported in a few cases before (32, 33); most of the cases with true hematoma are reported at the delayed phase after CABG (29–31), while most of the early cases are found to be pseudoaneurysms. Therefore, hematoma should be considered in the differential diagnoses of patients with early postoperative complications, as well. Also, the common sites of post-CABG hematoma are pericardial (29, 34) and very rarely retrosternal (like our case). The concurrency of retrosternal hematoma and TdP in our patient was a rare finding, and the resolution of this malignant arrhythmia by removal of hematoma was an important finding that has to be noted in future studies.

Conclusion

Retrosternal hematoma can form after CABG as an early postoperative complication. Arrhythmias may also occur or aggregate as a result of surgery. Co-occurrence of retrosternal hematoma and arrhythmia (drug-resistant TdP) after CABG has been noted in the present case, as a rare finding. Bearing in mind that TdP is a malignant arrhythmia and may result in fatal complications, it is important to find the exact cause of TdP occurrence, among the multiple risk factors, in order to find an appropriate treatment; many cases are refractory to conventional treatments. In the case presented here, the resolution of both post-CABG complications after the second surgery, removal of hematoma, suggested the possibility of a “cause and effect” relationship between hematoma and TdP. This finding has not been reported previously, and our finding suggest that physicians should consider surgical removal of hematoma for the treatment of similar cases, who develop resistant TdP. However, further studies are required to confirm this finding.

Data availability statement

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

Ethics statement

All procedures performed in the study were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration, and its later amendments or comparable ethical standards. 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

MS: Data curation, Investigation, Methodology, Resources, Writing – original draft, Writing – review & editing. MG: Data curation, Methodology, Writing – original draft, Writing – review & editing. ML: Data curation, Methodology, Writing – review & editing. SK: Data curation, Investigation, Methodology, Writing – review & editing. TD: 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.

References

1. Jayaraj JC, Davatyan K, Subramanian S, Priya J. Epidemiology of myocardial infarction. In: Pamukcu B, editors. *Myocardial Infarction*. 3rd ed. Rijeka: IntechOpen (2018). doi: 10.5772/intechopen.74768
2. Lawton JS, Tamis-Holland JE, Bangalore S, Bates ER, Beckie TM, Bischoff JM, et al. 2021 ACC/AHA/SCAI guideline for coronary artery revascularization: executive summary: a report of the American College of Cardiology/American Heart Association Joint Committee on clinical practice guidelines. *J Am Coll Cardiol.* (2022) 79:197–215. doi: 10.1016/j.jacc.2021.09.005
3. Durko AP, Budde RP, Geleijnse ML, Kappetein AP. Recognition, assessment and management of the mechanical complications of acute myocardial infarction. *Heart.* (2018) 104:1216–23. doi: 10.1136/heartjnl-2017-311473
4. Jawitz OK, Gulack BC, Brennan JM, Thibault DP, Wang A, O'Brien SM, et al. Association of postoperative complications and outcomes following coronary artery bypass grafting. *Am Heart J.* (2020) 222:220–8. doi: 10.1016/j.ahj.2020.02.002
5. Chiriac L, Rosulescu R. Arrhythmias and conduction disturbances after coronary artery bypass graft surgery. In: Tintoiu I, Underwood M, Cook S, Kitabata H, Abbas A, editors. *Coronary Graft Failure*. Switzerland: Springer, Cham (2016). p. 167–74.
6. Halkin A, Roth A, Lurie I, Fish R, Belhassen B, Viskin S. Pause-dependent torsade de pointes following acute myocardial infarction: a variant of the acquired long QT syndrome. *J Am Coll Cardiol.* (2001) 38:1168–74. doi: 10.1016/s0735-1097(01)01468-1
7. Lee G, Sanders P, Kalman JM. Catheter ablation of atrial arrhythmias: state of the art. *Lancet.* (2012) 380:1509–19. doi: 10.1016/s0140-6736(12)61463-9
8. Egbe AC, Connolly HM, McLeod CJ, Ammash NM, Niaz T, Yogeswaran V, et al. Thrombotic and embolic complications associated with atrial arrhythmia after Fontan operation: role of prophylactic therapy. *J Am Coll Cardiol.* (2016) 68:1312–9. doi: 10.1016/j.jacc.2016.06.056
9. Peretto G, Durante A, Limite LR, Cianflone D. Postoperative arrhythmias after cardiac surgery: incidence, risk factors, and therapeutic management. *Cardiol Res Pract.* (2014) 2014:615987. doi: 10.1155/2014/615987
10. Neira V, Enriquez A, Simpson C, Baranchuk A. Update on long QT syndrome. *J Cardiovasc Electrophysiol.* (2019) 30:3068–78. doi: 10.1111/jce.14227
11. Johnston J, Pal S, Nagele P. Perioperative torsade de pointes: a systematic review of published case reports. *Anesth Analg.* (2013) 117:559–64. doi: 10.1213/ANE.0b013e318290c380
12. Iguina MM, Smithson S, Danckers M. Incessant refractory polymorphic ventricular tachycardia after coronary artery bypass graft. *Cureus.* (2021) 13:e12752. doi: 10.7759/cureus.12752
13. Pasley T, Gillham M. The effect of cardiac surgery on the QT interval. Are patients at higher risk of torsades de pointes post cardiac surgery? *Heart Lung Circ.* (2014) 23:e16–e7. doi: 10.1016/j.hlc.2014.04.168
14. William J, Shembrey J, Quine E, Perrin M, Ridley D, Parameswaran R, et al. Polymorphic ventricular tachycardia storm after coronary artery bypass graft surgery: a form of 'angry purkinje syndrome'. *Heart Lung Circ.* (2023) 32:986–92. doi: 10.1016/j.hlc.2023.04.298
15. Kodaka M, Mori T, Ichikawa J, Ando K, Komori M. Refractory ventricular arrhythmias during aortic valve replacement and cardiac artery bypass requiring 16 attempts of electrical cardioversion: a case report. *JA Clin Rep.* (2020) 6:60. doi: 10.1186/s40981-020-00369-w
16. Wallace E, Howard L, Liu M, O'Brien T, Ward D, Shen S, et al. Long QT syndrome: genetics and future perspective. *Pediatr Cardiol.* (2019) 40:1419–30. doi: 10.1007/s00246-019-02151-x
17. Schwartz PJ, Woosley RL. Predicting the unpredictable: drug-induced QT prolongation, and torsades de pointes. *J Am Coll Cardiol.* (2016) 67:1639–50. doi: 10.1016/j.jacc.2015.12.063
18. Beach SR, Celano CM, Noseworthy PA, Januzzi JL, Huffman JC. QTc prolongation, torsades de pointes, and psychotropic medications. *Psychosomatics.* (2013) 54:1–13. doi: 10.1016/j.psym.2012.11.001
19. Niimi N, Yuki K, Zaleski K. Long QT syndrome and perioperative torsades de pointes: what the anesthesiologist should know. *J Cardiothorac Vasc Anesth.* (2022) 36:286–302. doi: 10.1053/j.jvca.2020.12.011
20. Iragavarapu T, Krishna K. Torsades De pointes complicating acute myocardial infarction, twisting the prognosis. *Indian Journal of Clinical Cardiology.* (2023) 4:208–14. doi: 10.1177/26324636231190247
21. Gowdak LHW, Bittencourt MS, Rochitte CE, Dallan LAO, César LAM. Coronary atherosclerotic plaque rupture following thoracic trauma: an uncommon cause of angina and ventricular tachycardia ("torsade de pointes"). *Clinics (Sao Paulo).* (2011) 66:1291–3. doi: 10.1590/s1807-59322011000700029
22. McCoy C, Miller JM, Tanawuttiwat T. A woman with recurrent torsade de pointes. *JAMA Cardiol.* (2023) 8:400–1. doi: 10.1001/jamacardio.2022.5094
23. Bottiglioni D, Monaco I, Santacroce R, Casavecchia G, Correale M, Guastaferro F, et al. Novel AKAP9 mutation and long QT syndrome in a patient with torsades des pointes. *J Interv Card Electrophysiol.* (2019) 56:171–2. doi: 10.1007/s10840-019-00606-y
24. Carpenter K, Ahmed I, Boland T. Torsades de pointes as a late complication of subarachnoid hemorrhage (P2. 287). *Neurology.* (2017) 88:P2.287. doi: 10.1212/WNL.88.16_supplement.P2.287
25. Wang K-C, Chen S-Y. Cerebellar hemorrhage presented as torsades de pointes. *Tungs' Med J.* (2022) 16:34–6. doi: 10.53106/207135922022061601007
26. Ibrahim R, Yadav S, Waqar S, Hermann JR, Sarwar A, Shah S. Superior vena Cava syndrome due to right anterior mediastinal hematoma: a case report. *Cureus.* (2022) 14:e26994. doi: 10.7759/cureus.26994

Publisher's note

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

Supplementary material

The Supplementary Material for this article can be found online at: <https://www.frontiersin.org/articles/10.3389/fcvm.2024.1331873/full#supplementary-material>

SUPPLEMENTARY FIGURE S1

The post-CABG ECG of the patient in the intensive care unit, which illustrates normal sinus rhythm with low voltage QRS, and bigeminal ventricular extrasystole (PVCs) with short coupling interval (R on T wave). doi: 10.1016/j.hlc.2023.04.298

27. Kanzaki H. Invisible hematoma causing shock after open-heart surgery: localized cardiac tamponade. *J Cardiol Cases*. (2014) 9:243–4. doi: 10.1016/j.jccase.2014.02.002

28. Helmy IM, Asbeutah AM, Elfiki IM, Arafa OE. A pictorial review on the role of 64-slice HD MDCT in detecting post CABG cardiothoracic complications. *Open Journal of Thoracic Surgery*. (2014) 4:48–58. doi: 10.4236/ojts.2014.42011

29. Floerchinger B, Camboni D, Schopka S, Kolat P, Hilker M, Schmid C. Delayed cardiac tamponade after open heart surgery—is supplemental CT imaging reasonable? *J Cardiothorac Surg*. (2013) 8:158. doi: 10.1186/1749-8090-8-158

30. Ohira S, Matsushita T, Masuda S. Late mediastinal hematoma presenting cardiac tamponade after re-do off-pump coronary artery bypass grafting in octogenarian; report of a case. *Kyobu Geka*. (2012) 65:1003–5.

31. Yamamoto T, Takahashi K, Tsukioka K, Kono T. Successful repair of chronic expanding hematoma after coronary artery bypass grafting by lower partial sternotomy approach; report of a case. *Kyobu Geka*. (2017) 70:863–6.

32. Ananthasubramaniam K, Jaffery Z. Postoperative right atrial compression by extracardiac hematoma: transesophageal echocardiographic diagnosis in the critically ill patient. *Echocardiography*. (2007) 24:661–3. doi: 10.1111/j.1540-8175.2007.00445.x

33. Grumann A, Baretto L, Dugard A, Morera P, Cornu E, Amiel J-B, et al. Localized cardiac tamponade after open-heart surgery. *Ann Thorac Cardiovasc Surg*. (2012) 18:524–9. doi: 10.5761/actcs.11.01855

34. Khan Z, Srour K, Khan M, Moustafa A, Javaid T. Anterior mediastinal hematoma and right sided hemothorax from leaking saphenous vein right coronary artery bypass graft aneurysm due to incomplete coiling masquerading as right lower lobe pneumonia. *Am J Respir Crit Care Med*. (2018) 197:A3445. doi: 10.1007/s11739-018-1847-5

35. Thomas SH, Behr ER. Pharmacological treatment of acquired QT prolongation and torsades de pointes. *Br J Clin Pharmacol*. (2016) 81:420–7. doi: 10.1111/bcpt.12726



OPEN ACCESS

EDITED BY

Martin Andreas,
Medical University of Vienna, Austria

REVIEWED BY

Philemon Gukop,
St George's University Hospitals NHS
Foundation Trust, United Kingdom
Massimo Baudo,
Lankenau Institute for Medical Research,
United States

*CORRESPONDENCE

Nina Sophie Pommert
✉ ninasophie.pommert@uksh.de

RECEIVED 20 January 2024

ACCEPTED 25 April 2024

PUBLISHED 20 May 2024

CITATION

Pommert NS, Puehler T, Voges I, Sellers S and Lutter G (2024) TMVR after TA-TAVR: a re-redo surgery—case report. *Front. Cardiovasc. Med.* 11:1373840. doi: 10.3389/fcvm.2024.1373840

COPYRIGHT

© 2024 Pommert, Puehler, Voges, Sellers and Lutter. This is an open-access article distributed under the terms of the [Creative Commons Attribution License \(CC BY\)](#). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

TMVR after TA-TAVR: a re-redo surgery—case report

Nina Sophie Pommert^{1,2*}, Thomas Puehler^{2,3}, Inga Voges⁴,
Stephanie Sellers^{5,6,7} and Georg Lutter^{1,2}

¹Department of Cardiac Surgery, University Medical Center Schleswig-Holstein, Campus Kiel, Kiel, Germany, ²DZHK (German Centre for Cardiovascular Research), Partner Site Hamburg/Kiel/Luebeck, Kiel, Germany, ³Department of Cardiac Surgery, University Medical Center Schleswig-Holstein, Campus Luebeck, Luebeck, Germany, ⁴Department of Congenital Heart Disease and Pediatric Cardiology, University Children's Hospital Kiel, Kiel, Germany, ⁵Centre for Cardiovascular Innovation, St Paul's and Vancouver General Hospital, Vancouver, BC, Canada, ⁶Cardiovascular Translational Laboratory, Providence Research & Centre for Heart Lung Innovation, Vancouver, BC, Canada, ⁷Centre for Heart Valve Innovation, St. Paul's Hospital, University of British Columbia, Vancouver, BC, Canada

Introduction: Transcatheter mitral valve replacement (TMVR) is a valuable treatment option in patients with severe mitral regurgitation. Prior transapical transcatheter aortic valve replacement (TA-TAVR) may complicate the procedure and is therefore considered a relative contraindication. In this case report, the authors describe the successful TMVR as a tertiary cardiac surgery and transapical redo procedure.

Case Summary: An 83-year-old male patient, suffering from dyspnoea and angina, was diagnosed with severe mitral valve regurgitation (MR). He had already undergone cardiac surgery in the form of coronary artery bypass grafting at the age of 64 and TA-TAVR at 79 years. After a failed attempt at mitral valve transcatheter edge-to-edge repair, he opted for TMVR. Pre-TMVR computed tomography simulation was used to analyse possible interactions between the prostheses and to predict the neo-left ventricular outflow tract (neo-LVOT). The operation was carried out without complications. There was no bleeding and the LV function remained unchanged. On MRI, the valves were perfectly aligned without any signs of paravalvular leakage or LVOT obstruction. The patient was discharged seven days postoperatively. At the one-year follow up, there was no need for rehospitalisation and the patient had clinically improved (from NYHA IV to II). Echocardiography demonstrated a mean transvalvular gradient of under 5 mmHg and no residual MR.

Conclusion: A redo transapical access for TMVR as a tertiary cardiac operation can be easily performed. Pre-operative CT suggested good alignment of the aortic and mitral valved stent which was confirmed postoperatively.

KEYWORDS

TAVR, TMVR, Tendyne, redo transapical access, case report

Introduction

Transcatheter mitral valve replacement (TMVR) has become a valuable treatment option in patients with severe mitral regurgitation (MR). Previous aortic valve replacement is considered a relative contraindication for the procedure due to the risk of left ventricular outflow tract (LVOT) obstruction and interactions between the two prostheses and anchoring mechanism. Moreover, in transapical redo surgery, complications at the access site may occur. In this case, the authors describe the successful TMVR as a tertiary cardiac surgery using the Tendyne® system.

Case presentation and diagnostic assessment

An 83-year-old male patient presented with dyspnoea and angina under cardiac decompensation (NYHA III). He had previously been treated with coronary artery bypass grafting for severe three-vessel disease at the age of 64 and transapical transcatheter aortic valve replacement (TA-TAVR, 29 mm S3) for aortic valve stenosis at 79 years. Coronary treatment was complemented by percutaneous coronary intervention and stenting of the PL branch, also at 79 years. Moreover, he was known to have arterial and pulmonary artery hypertension, chronic obstructive lung disease and peripheral artery disease with a history of bilateral femoropopliteal bypass surgery (Table 1). Before the start of the complaint, he lived at home independently.

On admission, blood pressure was elevated (180/100 mmHg); heart rate (71 bpm) and room air oxygen saturation (98%) were normal. Pulmonary auscultation revealed reduced breathing sounds, in line with pulmonary oedema in the thoracic x-ray. The ECG showed a normal frequency sinus rhythm with left axis deviation and bifascicular block. Echocardiography revealed severe secondary mitral valve regurgitation (MR) with an effective regurgitation orifice area (EROA) of 29 mm² with a vena contracta width of 7 mm, and a regurgitation volume of 50 ml. The left ventricular function was moderately impaired (ejection fraction 36%, left ventricular end diastolic volume 215 ml) and there was moderate tricuspid regurgitation. Laboratory analysis on admission revealed elevated of NT-proBNP (6,417 ng/L). Due to elevated troponin T with an increasing trend, coronary angiography was performed, excluding renewed coronary stenoses.

Therapeutic intervention

The patient was discussed by a multidisciplinary heart team and initially primed for mitral valve transcatheter edge-to-edge repair, but the procedure was not successful due to strong tethering of the posterior leaflet. Considering the high-risk profile

(STS-Score 7.5%, EuroScore II 35.4%) the patient opted for TMVR with the Tendyne® system.

Pre-procedural computed tomography (CT) simulation was used to determine the ideal access and exclude interactions between the mitral and aortic valve prosthesis. Special attention was paid to the neo-LVOT, which was predicted to have 10.6 mm (Figure 1A).

TMVR was performed under echocardiographic and fluoroscopic guidance, using the previous anterolateral thoracotomy for transapical access. The partially pulmonary adhesion was carefully dissected from the thoracic wall and the cardiac apex, revealing the pledges of the previous transapical procedure. For puncturing of the left ventricle with optimal angulation to the mitral position, a site posterolateral to the previous access was chosen. After apical pre-dilation with a 21F sheath, a 26F sheath was placed, through which the implantation was carried out without complications. Fluoroscopy by the end of the procedure confirmed optimal valve positioning (Figure 1B). The patient was extubated in the operating room and was admitted to normal ward on the first postoperative day.

Postoperative magnetic resonance imaging (MRI) showed optimal positioning of the prosthesis in the mitral anulus

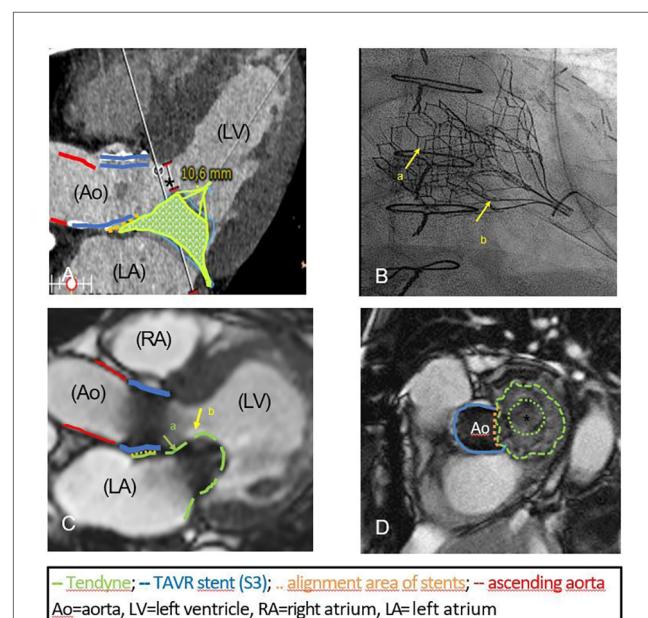


FIGURE 1

(A) Three-chamber view in CT simulation, predicting an end-diastolic neo-LVOT of 10.6 mm (*). The simulated alignment area of the aortic (29 mm S 3, blue line) and mitral prosthesis (green cage) is marked by the dotted orange line. The red line indicates the ascending aorta. (B) Fluoroscopy at the end of the procedure: 29 mm S 3 (arrow a) and the overlapping crown of the 29 mm Tendyne valve (low profile, arrow b) are fully deployed without interference. (C) Postprocedural MRI documenting the well-placed intra-annular position of Tendyne valve (arrow a) and its distal part targeting the anterior leaflet (hyperintense, arrow b, length 23.4 mm). The ascending aorta (red line), the cylinder of the S3 (blue line), the implanted Tendyne® stent (green line) and the overlapping area (dotted orange line) are highlighted. No LVOT-obstruction. (D) Optimal alignment (orange dotted line) of the S3 (blue line) and Tendyne prosthesis (green line) on postoperative MRI. The circular inner frame of the Tendyne valve with porcine pericardial leaflets (*) is mounted on a self-expanding outer frame.

TABLE 1 Patient timeline.

Timeline	
1999	Bilateral femoropopliteal bypass surgery
2003	Coronary artery bypass grafting (RIMA to LAD, SVG to PDA and PL)
09/2018	Cardiac decompensation NYHA IV
09/2018	PCI and stenting of PL branch
11/18	Transapical TAVR (29 mm S3)
06/22	Onset of angina and dyspnoea NYHA III-IV <ul style="list-style-type: none"> - Exclusion of relevant coronary stenoses - Diagnosis of severe MR
08/22	Failed TEER
09/22	TMVR (Tendyne 29 L)
09/22	Discharge home in good clinical condition
09/23	Follow-up at 12 months: good clinical condition, living independently at home, dyspnoea (NYHA II)

without any sign of paravalvular leakage, which was confirmed by echocardiography (Figures 1C,D). LV function remained moderately impaired. As predicted, there was no obstruction of the neo-LVOT. Haemoglobin was stable postoperatively with no need for transfusions.

The patient was discharged home seven days postoperatively. He subsequently participated in a three-week inpatient cardiac rehabilitation programme. There were no postoperative complications according to the MVARC criteria and no further inpatient admission was necessary in the year that followed. Echocardiography excluded paravalvular leakage or relevant transvalvular gradients (< 5 mmHg). LV function was only lightly impaired. The patient continued to live independently at home with improved exercise capacity (NYHA II).

Discussion

Multi-valvular disease in elderly, multimorbid patients is an important issue to deal with. With more than 1,700 procedures performed worldwide, TMVR with the Tendyne® system (Abbott) is an emerging treatment strategy for severe mitral valve regurgitation in high-risk patients.

As it is potentially fatal, LVOT obstruction is of major concern in patients with TMVR. Implantation of the prosthesis leads to the formation of the so called neo-LVOT, confined by the native anterior mitral leaflet fixed in an opened position, the valve stent and the septum (1). LVOT obstruction may lead to acute hemodynamic deterioration or chronic heart failure due to an increased left ventricular afterload (1, 2). As a consequence, pre-procedural computed tomography simulation and measurement of the predicted LVOT has gained in importance (1).

An existing aortic valve prosthesis may add complexity as it influences aortomitral angulation. Moreover, LV hypertrophy as a consequence of long-standing aortic valve stenosis may narrow the LVOT. We therefore relied on CT simulation in this complex case to visualise the planned implantation and ensure there was an adequate LVOT area.

The feasibility of TMVR in the presence of a surgical or transcatheter aortic valve prosthesis has previously been described previously (3, 4). Similarly, simultaneous transapical implantations of an aortic and mitral valve prosthesis has been reported (5). However, in both aortic and mitral valve surgery, little is known about transapical re-intervention. Despite the fear of apical fragility, individual reports describe good results of redo transapical TAVR within a week, three or seven years after the initial procedure (6–8). As in our case, postoperative adhesions were easily controllable and the apical tissue was rated unexpectedly normal (6). In the cases mentioned of redo transapical TAVR, the same apical access site was used. For optimal angulation in our patient, we had to use a new apical access site posterolateral of the previous position. Transapical access may cause myocardial damage and scarring. However, the existence of two adjacent accesses did not lead to restriction of the left ventricular function in our patient. He showed moderately impaired LV function in both pre- and postprocedural

echocardiography and MRI (ejection fraction 36% pre- and 37% post-procedurally). There was even an improvement to a lightly impaired left ventricular function in the one-year follow up.

In conclusion, this case demonstrates that a redo transapical access for TMVR as a tertiary cardiac procedure can be easily performed with good clinical results. Accurate pre-operative screening is crucial.

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 studies involving humans because we presented our project to our ethics committee, who confirmed that an ethical statement on a case report is not necessary if patient informed consent is available. 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 participant/patient(s) for the publication of this case report.

Author contributions

NP: Writing – original draft. TP: Conceptualization, Supervision, Visualization, Writing – review & editing. IV: Data curation, Investigation, Visualization, Writing – review & editing. SS: Supervision, Writing – review & editing. GL: Conceptualization, Data curation, Funding acquisition, Methodology, Resources, Supervision, Validation, Visualization, Writing – review & editing.

Funding

The author(s) declare that financial support was received for the research, authorship, and/or publication of this article.

We acknowledge financial support by Land Schleswig-Holstein within the funding programme Open Access Publikationsfonds.

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.

Publisher's note

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated

organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

References

1. Reid A, Ben Zekry S, Turaga M, Tarazi S, Bax JJ, Wang DD, et al. Neo-LVOT and transcatheter mitral valve replacement: expert recommendations. *JACC Cardiovasc Imaging*. (2021) 14(4):854–66. doi: 10.1016/j.jcmg.2020.09.027
2. Yoon SH, Bleiziffer S, Latib A, Eschenbach L, Ancona M, Vincent F, et al. Predictors of left ventricular outflow tract obstruction after transcatheter mitral valve replacement. *JACC Cardiovasc Interv*. (2019) 12(2):182–93. doi: 10.1016/j.jcin.2018.12.001
3. Cheung A, Webb J, Schaefer U, Moss R, Deuschl FG, Conradi L, et al. Transcatheter mitral valve replacement in patients with previous aortic valve replacement. *Circ Cardiovasc Interv*. (2018) 11(10):e006412. doi: 10.1161/CIRCINTERVENTIONS.118.006412
4. Taramasso M, Sorajja P, Dahle G, et al. Transapical transcatheter mitral valve implantation in patients with prior aortic valve replacement: a feasibility report. *EuroIntervention*. (2021) 17(3):257–9. doi: 10.4244/EIJ-D-19-00947
5. Elkharbony A, Delago A, El-Hajjar M. Simultaneous transapical transcatheter aortic valve replacement and transcatheter mitral valve replacement for native valvular stenosis. *Catheter Cardiovasc Interv*. (2016) 87(7):1347–51. doi: 10.1002/ccd.26078
6. Ferrari E, Locca D, Marcucci C, Jeanrenaud X. Urgent reoperative transapical valve-in-valve shortly after a transapical aortic valve implantation. *Eur J Cardiothorac Surg*. (2014) 46(4):748–50. doi: 10.1093/ejcts/ezt552
7. Kiefer P, Seeburger J, Chu MW, Ender J, Vollroth M, Noack T, et al. Reoperative transapical aortic valve implantation for early structural valve deterioration of a SAPIEN XT valve. *Ann Thorac Surg*. (2013) 95(6):2169–70. doi: 10.1016/j.athoracsur.2012.10.072
8. Ricciardi G, Cavallotti L, Alamanni F, Roberto M. Reoperative transapical transcatheter aortic valve implantation for a degenerated biological valve. *JTCVS Tech*. (2020) 4:118–20. doi: 10.1016/j.xjtc.2020.10.037



OPEN ACCESS

EDITED BY

Giuseppe Gatti,
Azienda Sanitaria Universitaria Giuliano
Isontina, Italy

REVIEWED BY

Lukas Stolz,
Ludwig Maximilian University of Munich,
Germany
Mustafa Kurkluoglu,
Northwestern Memorial Hospital,
United States

*CORRESPONDENCE

Jinfeng Zhou
✉ Zhoujinfengaa@sina.com

[†]These authors have contributed equally to
this work and share first authorship

RECEIVED 07 April 2024

ACCEPTED 15 July 2024

PUBLISHED 05 August 2024

CITATION

Wang M, Ji F and Zhou J (2024) Case Report:
Left atrial dissection after mitral valve
replacement: intraoperative management
under TEE guidance.
Front. Cardiovasc. Med. 11:1413713.
doi: 10.3389/fcvm.2024.1413713

COPYRIGHT

© 2024 Wang, Ji and Zhou. This is an open-
access article distributed under the terms of
the [Creative Commons Attribution License
\(CC BY\)](#). The use, distribution or reproduction
in other forums is permitted, provided the
original author(s) and the copyright owner(s)
are credited and that the original publication in
this journal is cited, in accordance with
accepted academic practice. No use,
distribution or reproduction is permitted
which does not comply with these terms.

Case Report: Left atrial dissection after mitral valve replacement: intraoperative management under TEE guidance

Mengyan Wang[†], Fucheng Ji[†] and Jinfeng Zhou*

Department of Anesthesiology, Qilu Hospital (Qingdao), Cheeloo College of Medicine,
Shandong University, Qingdao, China

Left atrial dissection (LatD) is a very rare complication of cardiac surgery, but it is relatively more common in mitral valve surgery. Transesophageal echocardiography (TEE) plays an important role in timely detection of LatD and accurate assessment of the condition, which are key factors in determining the patient's prognosis. There are two different treatment options for patients with or without circulatory crisis caused by dissection hematoma, namely surgical management and conservative treatment. In this report, we used TEE to quickly detect the cause and severity of LatD, which assisted the surgeon in making appropriate surgical decisions. The patient was successfully surgically treated for LatD.

KEYWORDS

atrial dissection, mitral valve replacement, left atrioventricular groove, TEE, surgical intervention

1 Introduction

Our patient was a 64-year-old woman who unexpectedly developed left atrial dissection (LatD) during mitral valve replacement surgery. Perioperative LatD is mostly iatrogenic. In this case, transesophageal echocardiography (TEE) was used to discover the site of injury that caused LatD, which is different from that in previously reported cases. Based on the evidence discovered by TEE, the surgeon was able to treat the patient successfully.

2 Case report

A 64-year-old woman was admitted to our cardiac surgery clinic with a complaint of chest tightness and dyspnea, which started 5 months ago without any apparent trigger. The symptoms worsened after exercise, accompanied by profuse sweating, dizziness, headache, abdominal bloating, and pain. She underwent TTE at a local hospital which revealed mitral valve prolapse (P2, P3 areas), severe mitral regurgitation, and left atrial enlargement. Because conservative treatment was ineffective, she was admitted to our hospital and referred for surgical intervention.

The laboratory test results were unremarkable. Coronary angiography showed no evidence of obstructive disease.

Following routine cardiac general anesthetic procedures, TEE confirmed severe eccentric mitral regurgitation owing to prolapse of the middle scallop of the posterior

mitral leaflet along with light tricuspid regurgitation and pulmonary valve regurgitation. The patient's left ventricular ejection fraction (LVEF) was normal (65%). A median sternotomy was performed, followed by standard right atrial and atrial septum access. Mitral valve replacement involved continuous suturing with 2-0 prolene thread, placement of a size 29 bioprosthetic valve, and ligation of the left atrial appendage with double 10-0 sutures. Subsequently, the aortic root was thoroughly deaired under TEE guidance, the aortic clamp was released, and the heart spontaneously started to re-beat. Unexpectedly, during the hemostasis process, the patient's blood pressure gradually decreased, requiring an increase in the dosage of adrenaline and noradrenaline to maintain an appropriate level. At this time, there was no obvious blood loss spots; we also excluded the possible use of medications such as protamine or antibiotics that could have triggered allergic reactions. To determine the cause of hypotension, we conducted an investigation using TEE and discovered a hematoma on the posterior wall of the left atrium, measuring approximately 3 × 4 cm in diameter (Figure 1). This hematoma was rapidly expanding and gradually compressing the mitral valve annulus.

Given this emergent situation, cardiopulmonary bypass was reinitiated to resolve the circulation crisis. The results of TEE monitoring showed that the left atrial dissection hematoma was compressing the left ventricular inflow tract, which accelerated blood flow through the mitral valve, causing mitral valve relative stenosis. In addition, TEE found pulsatile blood flow in the hematoma at the left atrioventricular groove on the posterior wall

of the left atrium after the second cardiopulmonary bypass (Figure 2), suggesting that this blood flow was related to the left coronary circumflex branch. However, the relationship between the left circumflex branch and the dissection could not be determined. Because the criminal's blood flow (blood vessels) was located in the posterior atrioventricular groove and could not be explored through a surgical incision, we ultimately decided to incise the left atrial endocardium to drain the decompression hematoma through the left atrium (Figures 3, 4). The specific method applied was to reopen the right atrium and atrial septum along the original surgical incision. After confirming the correct position of the artificial valve and the integrity of the left atrial endocardium, the latter was incised for drainage and decompression, and the atrial septum and right atrium were resutured. The patient was admitted to the ICU for further management and discharged home after 16 days.

Follow-up transthoracic ultrasound at 15 and 25 days after surgery both showed heterogeneous echogenicity on the left atrial posterior wall, measuring approximately 26 mm × 12 mm, with no significant change compared to before. The ejection fraction remained at 63%.

3 Discussion

Left atrial dissection is defined as a false, blood-filled cavity or lumen from the mitral annular area to the left atrial free wall or interatrial septum that creates a new chamber with or without

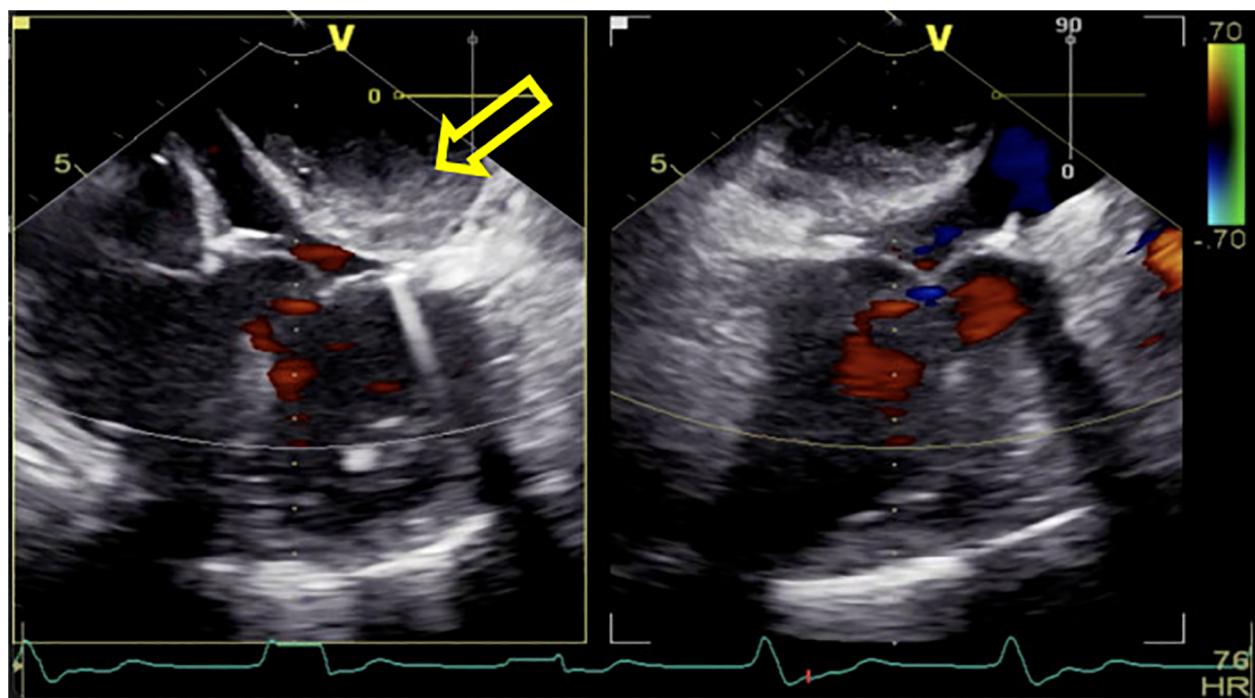


FIGURE 1

After being weaned from CPB, a rapidly increasing left atrial dissection hematoma (indicated by the yellow arrow) was visible in the posterior atrioventricular groove of the left atrial posterior wall. It gradually compressed the mitral valve annulus, causing relative stenosis of the left ventricular inflow tract.

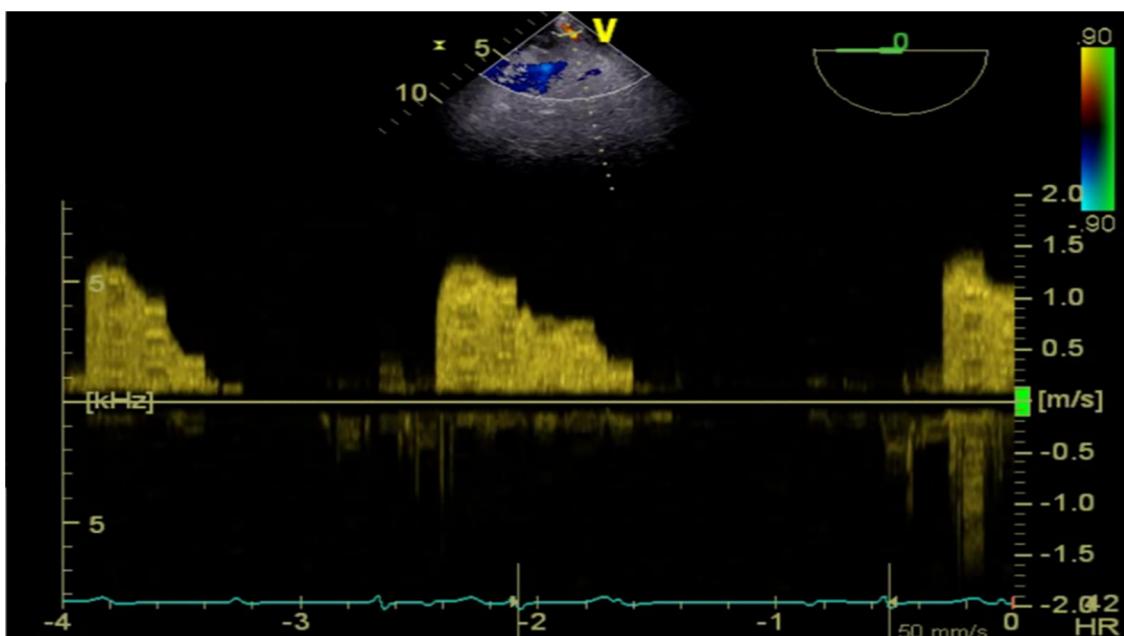


FIGURE 2
TEE detected pulsatile blood flow in the hematoma located at the left atrioventricular groove on the posterior wall of the left atrium.

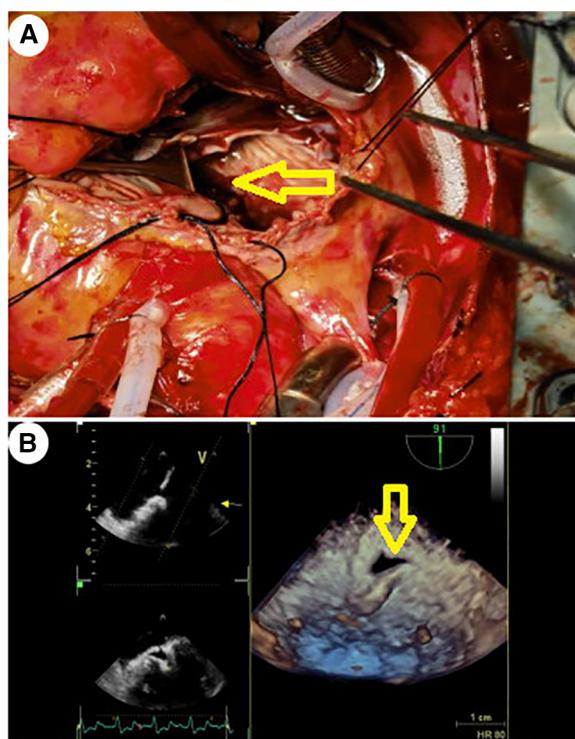


FIGURE 3
The yellow arrow indicates the site of incision and drainage. (A) Surgical treatment method: After confirming the integrity of the left atrial endocardium and the correct position of the artificial valve, the left atrial endocardium was incised for decompression and drainage. (B) TEE 3D reconstruction detected the drainage port of the left atrial endocardium incision for decompression.

communications into the true left atrium (1). LatD is a rare complication of cardiac surgery, with reported incidence rates ranging from 0.16% to 0.84%, which is traditionally associated with mitral valve surgery. However, with the development of percutaneous coronary intervention (PCI) in recent years, the number of cases reported after radiofrequency ablation and PCI has also slightly increased (2). The reasons for LatD caused by different surgeries are different, such as the atrioventricular groove damage (3) during mitral valve surgery, atrial wall injury from ablation procedures (4), and coronary artery perforation during PCI (5). Genoni et al. (3) claimed that the most likely source of LatD hematoma during mitral valve surgery was left ventricular arterial blood. Intense traction of sutures in weak tissue can cause bleeding from the atrioventricular groove, which is not directed outward toward the pericardial cavity, rather toward the atrium itself, leading to spread of tissue and formation of a cavity.

In this report, we discuss a case of LatD that occurred after mitral valve surgery, but it had a unique presentation compared to previous reports. In previous reports, most cases of atrial dissection after mitral valve surgery were caused by surgical operation leading to damage to the left atrial endometrium. In this case, the LatD occurred during the valve annulus suturing process, especially when the suture needle in the posterior annulus was too deep, which damaged the coronary branches in the atrioventricular groove.

The overall mortality rate for left atrial dissection is reportedly 13.8% (6), emphasizing the importance of timely detection and accurate diagnostic assessment. Continuous monitoring through TEE plays a crucial role in both diagnosis and management. Almost all reports of LatD in the past two decades have relied on

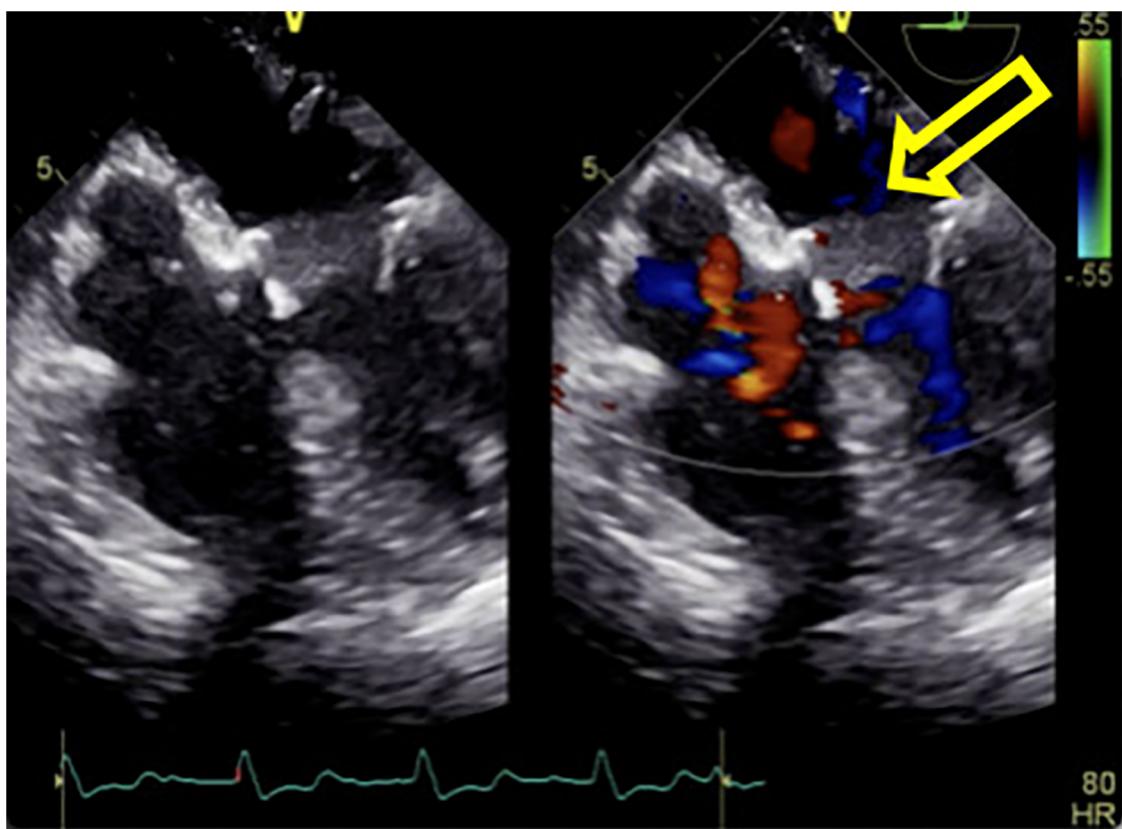


FIGURE 4

The hematoma on the posterior wall of the left atrium significantly decreased in size after incision of the left atrial endocardium for decompression and drainage.

TEE monitoring. Before the TEE era, LatD could only be detected through intraoperative general examination or occasionally, by autopsy (7, 8). Nowadays, TEE is the first-choice diagnostic method, and in some cases, the origin and extent of LatD are clearly demonstrated by TEE, with duplication of the left atrial wall and vast movement of the dissected wall (9). The clinical application of 3D TEE can accurately evaluate the anatomical factors causing hemodynamic instability in LatD, such as the degree of compression on the left atrium and pulmonary vein opening, obstruction of the left ventricular inflow tract, and determination of whether there is blood flow communicating the true and false lumens that can cause a decrease in cardiac output and heart failure. Using continuous TEE monitoring of the mass during protamine administration, the temporal relationship between reversal of heparin and the changing appearance of the mass from mostly fluid-filled hypoechoic mass to a hyperechoic mass as the blood coagulated could be clearly visualized (10). Reliable TEE evaluation can help surgical teams make the correct treatment decisions immediately, block further deterioration of patient circulation status, and avoid the risk of postoperative secondary thoracotomy or even death. For unexpected left atrial hematoma, treatment decisions are mainly based on the stability of the patient's hemodynamics. However, there are numerous uncertain factors in the progression of left atrial dissection. Even for patients who choose conservative treatment for current stable

hemodynamics, close monitoring should be carried out, especially continuous TEE monitoring (11).

In unstable patients, surgical treatment is the top priority. The purpose of surgical treatment is to eliminate hematoma, relieve compression, close the false lumen, and close the entrance (12). There are two methods for surgical treatment, one is internal drainage (3, 13, 14) and the other is entry closure (15–17) (i.e., by suturing the false cavity or tear layer). However, not all dissected hematomas' origins can be accurately located. Therefore, internal drainage often becomes the first choice for quickly alleviating hemodynamic collapse. In Genoni et al.'s (3) technique, internal drainage was applied to ensure drainage of a dissected left atrial wall into the right atrium. Hereby, the cavity of the left atrium is restored, while preventing systemic embolization and rupture by further increase of intracavitory pressure. Matteucci et al. (14) reported a new internal drainage method called atrial fenestration. In this case, during the extracorporeal circulation, owing to low blood pressure and laminar flow, a small-volume hematoma starts begins to develop. Additionally, because of the insufficient filling of the heart and the less standardized ultrasound section, any abnormalities that had already occurred can be easily overlooked.

Following the extracorporeal circulation stop, as the blood pressure increases, laminar flow shifts to pulsatile flow. Accelerated blood flow into the dissected layer rapidly enlarges

the hematoma on the left atrium's posterior wall. This significantly obstructs the left ventricular inflow tract, with Doppler indicating a narrowed mitral valve orifice area of 1.5 cm^2 , equivalent to moderate mitral stenosis. Subsequently, the team opted for surgical intervention to counter the LatD's severe impact on circulation. They reestablished extracorporeal circulation and alleviated pressure by incising and draining the tense area on the left atrium's endocardial posterior wall. This surgical approach effectively minimized cardiac damage and shortened the extracorporeal circulation time for the patient.

4 Conclusion

TEE monitoring assists anesthesiologists gain a deeper understanding of the structural and functional aspects of the heart during surgery. It serves to facilitate efficient communication between anesthesiologists and surgeons. TEE monitoring serves as a crucial foundation for surgical decision-making. It allows for real-time, comprehensive assessment of the structure and function of the heart, enabling timely diagnosis of the causes of hemodynamic instability. In this case, reliable TEE monitoring facilitated the timely identification of the circulatory instability trigger, enabling the surgical team to make informed decisions and avoid unplanned reoperations. The case underscores the significance of incorporating TEE throughout the cardiac surgery process, emphasizing its role in providing timely and accurate monitoring information.

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 was obtained from the

References

1. Gallego P, Oliver JM, González A, Domínguez FJ, Sanchez-Recalde A, Mesa JM. Left atrial dissection: pathogenesis, clinical course, and transesophageal echocardiographic recognition. *J Am Soc Echocardiogr.* (2001) 14(8):813–20. doi: 10.1067/mje.2001.113366
2. Cereda AF, De Luca F, Lanzone AM, Cottini M, Pastori L, Sangiorgi G. Case report and systematic review of iatrogenic left atrial dissection in different cardiovascular specialties: a common treatment for an uncommon complication? *Catheter Cardiovasc Interv.* (2020) 95(1):E30–6. doi: 10.1002/ccd.28356
3. Genoni M, Jenni R, Schmid ER, Vogt PR, Turina MI. Treatment of left atrial dissection after mitral repair: internal drainage. *Ann Thorac Surg.* (1999) 68:1394–6. doi: 10.1016/S0003-4975(99)00709-2
4. Kashou AH, DeSimone CV, Asirvatham SJ, Kapa S. Left atrial dissection as a trigger for recurrent atrial fibrillation. *HeartRhythm Case Rep.* (2020) 6(6):329–33. doi: 10.1016/j.hrcr.2020.02.011
5. Wang S, Weng J, He F, Qian X, Liu Y, Chen H. Surgical treatment of left atrial dissection caused by percutaneous coronary intervention. *Cardiovasc J Afr.* (2022) 33(4):220–4. doi: 10.5830/CVJA-2021-045
6. Fukuhara S, Dimitrova KR, Geller CM, Hoffman DM, Ko W, Trambulla RF. Left atrial dissection: etiology and treatment. *Ann Thorac Surg.* (2013) 95:1557–62. doi: 10.1016/j.athoracsur.2012.12.041
7. Choi JH, Kang JK, Park KJ, Jung JW, Choi SY, Yoon MH, et al. Images in cardiovascular medicine. Spontaneous left atrial dissection presenting as pulmonary edema. *Circulation.* (2005) 111(22):e372–3. doi: 10.1161/CIRCULATIONAHA.104.477463
8. Lang RM, Addetia K, Narang A, Mor-Avi V. 3-dimensional echocardiography: latest developments and future directions. *JACC Cardiovasc Imaging.* (2018) 11(12):1854–78. doi: 10.1016/j.jcmg.2018.06.024

individual(s) for the publication of any potentially identifiable images or data included in this article.

Author contributions

MW: Data curation, Writing – original draft, Writing – review & editing. FJ: Supervision, Writing – review & editing. JZ: Conceptualization, Resources, Supervision, Writing – review & editing.

Funding

The author(s) declare financial support was received for the research, authorship, and/or publication of this article. The work was funded by the Qingdao Key Health Discipline Development Fund (QDZDZK2022094).

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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

Supplementary material

The Supplementary Material for this article can be found online at: <https://www.frontiersin.org/articles/10.3389/fcvm.2024.1413713/full#supplementary-material>

9. Martínez-Sellés M, García-Fernandez MA, Moreno M, Bermejo J, Delcán JL. Echocardiographic features of left atrial dissection. *Eur J Echocardiogr.* (2000) 1 (2):147–50. doi: 10.1053/euje.2000.0020
10. Tolpin DA, Collard CD, Thomas Z, Pan W. Left atrial dissection associated with pulmonary vein cannulation. *Anesth Analg.* (2009) 109:1409–12. doi: 10.1213/ANE.0b013e3181b7c508
11. Morishita A, Katahira S, Hoshino T, Hanzawa K, Tomioka H. Rapidly vanishing left atrial dissection following mitral valve replacement: a case report. *J Cardiothorac Surg.* (2020) 15(1):73. doi: 10.1186/s13019-020-01112-3
12. Kasai M, Inoue Y, Suzuki S, Suzuki R. Acute left atrial dissection during redo mitral valve replacement. *J Card Surg.* (2016) 31(8):521–2. doi: 10.1111/jocs.12780
13. Schmid ER, Schmidlin D, Jenni R. Left atrial dissection after mitral valve reconstruction. *Heart.* (1997) 78:492. doi: 10.1136/hrt.78.5.492
14. Matteucci M, Torchio F, Messina C, Inzigneri G, Severgnini P, Musazzi A. Intraoperative left atrial dissection following mitral valve surgery: report of a case treated surgically. *J Card Surg.* (2022) 37(12):5545–7. doi: 10.1111/jocs.17087
15. Maeda K, Yamashida C, Shida T, Okada M, Nakamura K. Successful surgical treatment of dissecting left atrial aneurysm after mitral valve replacement. *Ann Thorac Surg.* (1985) 39(4):382–4. doi: 10.1016/S0003-4975(10)62639-2
16. Sekino Y, Sadahiro M, Tabayashi K. Successful surgical repair of left atrial dissection after mitral valve replacement. *Ann Thorac Surg.* (1996) 61:1528–30. doi: 10.1016/0003-4975(95)01157-9
17. Goda T, Ishii K, Shiiya N, Oba J, Matsui Y, Yasuda K. Acute dissection of the interatrial septum after re-replacement of the mitral valve: a case report. *J Jpn Assoc Thorac Surg.* (1994) 42:1092–5.



OPEN ACCESS

EDITED BY

Giuseppe Gatti,
Azienda Sanitaria Universitaria Giuliano
Isontina, Italy

REVIEWED BY

Ilaria Franzese,
Cattinara Hospital, Italy
Jayant Shyam Jainandunsing,
University Medical Center Groningen,
Netherlands

*CORRESPONDENCE

Rodrigo Estévez-Loureiro
✉ roiestevez@hotmail.com

RECEIVED 12 May 2024

ACCEPTED 05 August 2024

PUBLISHED 21 August 2024

CITATION

Caneiro-Queija B, Guerreiro CE, Echarte-Morales J, Estévez-Loureiro R, Barreiro-Pérez M, González-Ferreiro R, Estévez-Cid F, Legarra JJ, Baz JA and Íñiguez-Romo A (2024) Left ventricular outflow tract obstruction after transcatheter mitral valve replacement: a case report with a multifaceted approach. *Front. Cardiovasc. Med.* 11:1431639. doi: 10.3389/fcvm.2024.1431639

COPYRIGHT

© 2024 Caneiro-Queija, Guerreiro, Echarte-Morales, Estévez-Loureiro, Barreiro-Pérez, González-Ferreiro, Estévez-Cid, Legarra, Baz and Íñiguez-Romo. This is an open-access article distributed under the terms of the [Creative Commons Attribution License \(CC BY\)](#). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

Left ventricular outflow tract obstruction after transcatheter mitral valve replacement: a case report with a multifaceted approach

Berenice Caneiro-Queija^{1,2}, Claudio E. Guerreiro^{1,2},
Julio Echarte-Morales^{1,2}, Rodrigo Estévez-Loureiro^{1,2*},
Manuel Barreiro-Pérez^{1,2}, Rocío González-Ferreiro^{1,2},
Francisco Estévez-Cid³, Juan José Legarra³, Jose Antonio Baz^{1,2}
and Andrés Íñiguez-Romo^{1,2}

¹Department of Cardiology, Álvaro Cunqueiro University Hospital, Vigo, Spain, ²Cardiovascular Research Group, Department of Cardiology, Álvaro Cunqueiro University Hospital, Fundación Biomédica Galicia Sur, Servizo Galego de Saude, University of Vigo, Vigo, Spain, ³Department of Cardiovascular Surgery, Álvaro Cunqueiro University Hospital, Vigo, Spain

An 83-year-old woman was admitted to our center because of heart failure. Transthoracic echocardiography revealed severe mitral annular calcification resulting in a double mitral valve lesion. After discussion by the heart team, transcatheter mitral valve replacement with Tendyne (Abbott Structural, Santa Clara, CA, USA) was performed. Despite having a predicted neo-left ventricular outflow tract (LVOT) above the cut-off value, the patient developed clinically significant LVOT obstruction (LVOTO) refractory to medical treatment. This situation is often treated before the intervention, and dealing with LVOTO afterward can be challenging. After taking the patient's anatomy into consideration, we decided to perform alcohol septal ablation. Applying a combined strategy of medical treatment and intervention led to success. In this case report, we discuss this event and the strategies available for preventing and managing the condition.

KEYWORDS

Tendyne, left ventricular outflow tract obstruction, transcatheter mitral valve replacement, mitral annular calcification, alcohol septal ablation (ASA)

Introduction

According to recent data from the Euro Heart Survey, degenerative valvular heart disease is highly prevalent (1). Mitral annular calcification (MAC) is a degenerative age-dependent process leading to mitral regurgitation (MR) or mitral stenosis (MS). MAC is linked to cardiovascular risk factors, the female gender, and chronic kidney disease (2). Although surgery has been the gold standard treatment for mitral valve disease, patients with MAC have been associated with an increased risk of cardiac rupture at the atrioventricular junction, perivalvular leaks, or circumflex artery injury (3). Transcatheter mitral valve replacement (TMVR) techniques have emerged in recent years to overcome the challenges of MAC treatment. In this regard, the Tendyne valve (Abbott Structural, Santa Clara, CA, USA) has proved to be a feasible option in MAC

patients (4). Nonetheless, several procedural concerns need to be taken into consideration, such as left ventricular outflow tract obstruction (LVOTO) or paravalvular leak (PVL) (5).

Case description

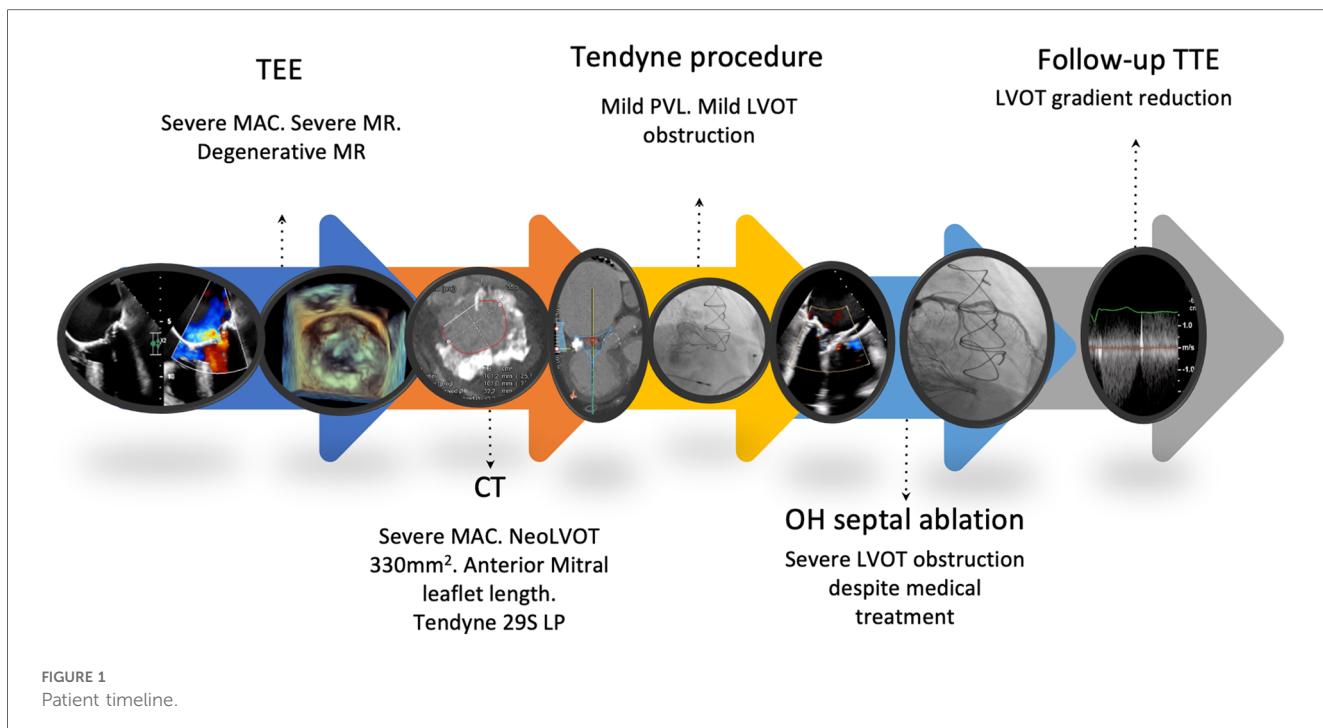
An 83-year-old woman was admitted to our center because of overt heart failure (HF). Her medical background revealed arterial hypertension, dyslipidemia, and obesity as cardiovascular risk factors. The patient also had a history of atrial fibrillation (AF). In 2012, she underwent cardiac surgery due to degenerative aortic stenosis, and a 21-mm aortic bioprosthesis (St. Jude Medical, Minnesota, USA) was implanted.

During the diagnostic work-up, echocardiography showed mild aortic bioprosthetic dysfunction, severe MAC resulting in a double mitral lesion (moderate MS and severe MR), and severe tricuspid regurgitation. The left ventricular ejection fraction (LVEF) was preserved with significant concentric hypertrophy (LV mass of 172 g/m² and septal thickness of 15 mm) (Supplementary Figure 1A). Coronary angiography evidenced chronic total occlusion of the mid-left anterior descending artery. Right heart catheterization showed a decreased cardiac output (2.2 L/min) with significant combined pulmonary hypertension (mean pulmonary artery pressure 43 mmHg, with pulmonary capillary wedge pressure 21 mmHg). Patient symptoms were mainly due to mitral valve disease. Due to the recording of high-risk scores (EuroSCORE II 15%, with society of thoracic surgeons (STS) score of 10.8% for mortality and 30% for morbidity), the heart team considered the patient to be at extreme risk for conventional intervention and decided to evaluate her for TMVR.

Transesophageal echocardiography (TEE) revealed a degenerated aortic bioprosthesis with mild-to-moderate regurgitation and a degenerative MR with evidence of chordal rupture, severe MR, severe MAC, and an aortic-mitral angle (AMA) of 135° (Supplementary Figure 1B). A computed tomography (CT) scan was also performed, with an MAC score (6) of 7 points and a predicted systolic NeoLVOT of 330 mm² (Supplementary Figure 2). A 29S LP Tendyne was thus deemed appropriate for the anatomy of the patient (Supplementary Video 1).

The procedure was performed under general anesthesia through a left mini-thoracotomy using a transapical approach with three-dimensional (3D) TEE imaging guidance. A standard 0.035-inch wire was inserted into the left atrium, and a balloon tip catheter was advanced to the left atrium to ensure a free chord trajectory. A 14-Fr sheath was inserted in the apex and the valve was pre-dilated with a 22 mm balloon to ensure calcium expansion (Supplementary Video 1). Then, a 34-Fr sheath was placed over the wire into the left atrium and the 29S LP Tendyne was delivered through the sheath and partially deployed in the left atrium, until the outer valve expanded to approximately 85% of the final size (Supplementary Video 2). The outer stent was aligned with the straight edge oriented anteriorly against the aortic-mitral continuum through device rotation under TEE guidance. The delivery sheath was retracted to deploy the remainder of the prosthesis in an intra-annular position. Finally, the length and tension of the tether were adjusted to optimize MR reduction and minimize the risk of device displacement. After the procedure, a mild posterior PVL was identified (Supplementary Video 3), without significant LVOTO (Supplementary Figure 2).

Days after the procedure, the patient exhibited poor AF rate control and HF. Transthoracic echocardiography (TTE) showed



LVOTO due to contact between the Tendyne medial frame and the septal wall (Supplementary Video 4), with an initial pressure gradient of 45 mmHg. Medical treatment for the LVOTO was initially established. Beta-blockers were up-titrated according to the clinical symptoms and tolerance. The patient improved, and TTE was performed, with an LVOT gradient of 35 mmHg before discharge. However, the patient was re-admitted with HF symptoms 18 days after discharge and repeated severe LVOTO (45 mmHg was noted on TTE). After carefully reviewing the patient's history, the heart team agreed to first implant a pacemaker, and then perform alcohol septal ablation (ASA) with a 1.25 mm over-the-wire balloon in the second septal perforation artery (Supplementary Video 5). Control TTE performed 7 weeks after ASA showed a LVOT gradient of 12 mmHg (Supplementary Figure 3). At present, 6 months after the procedure, the patient presents NYHA functional class II, without signs of HF and with no further hospital admissions (Figure 1).

Discussion

The present case report highlights the challenges associated with the transcatheter treatment of complex valvular heart disease and the broad range of interventional options we currently have to deal with them. Severe MAC is an uncommon condition, although it significantly increases the risk of mitral valve dysfunction with regurgitation and/or stenosis. Not only the increased patient age and related comorbidities, but also the associated anatomical features highlight the technical complexity of addressing this disease, which is linked to a greater risk of complications such as PVL, circumflex artery injury, disruption of the atrioventricular groove (7), conduction disturbances, and patient–prosthesis mismatch (8).

The current reported case corresponded to a high surgical risk patient of advanced age, with previous open-heart surgery and with isolated post-capillary pulmonary hypertension. To minimize the procedural risk, a stepwise approach should be taken into consideration. First, these cases require thorough discussion within a multidisciplinary heart team specialized in valve management, to properly individualize care, balancing the risk and benefit of an invasive strategy. Second, pre-procedural planning with multimodality imaging is pivotal, involving not only echocardiography but particularly also CT to assess the valvular anatomy and the extent and distribution of MAC—including the involvement of other adjacent structures—and to predict device implantation and its potential complications such as LVOTO. Initially, we estimated the LVOT-box area as described previously (9) to predict the risk of LVOTO, obtaining an area of 430 mm², thus we continued the Tendyne screening process. Taking into account the CT features of mitral annulus calcium thickness, distribution, and trigone and leaflet involvement, the MAC score was seen to be 7. According to Guerrero et al., this score is associated with a 5.86-fold increase in the odds of valve embolization/migration when using a balloon-expandable valve for valve-in-mitral annular calcification (ViMAC) transcatheter aortic valve replacement (TAVR) (6).

Also, mitral valve area (MVA) was close to 750 mm², which fits a dedicated mitral prosthesis better rather a TAVR in mitral position. In addition, the measured NeoLVOT was 330 mm², which was clearly in the theoretical safety zone, above the cut-off value of 189.4 mm². This threshold demonstrated a sensitivity of 100% and a specificity of approximately 97% in predicting a post-TMVR increase in LVOT gradient of 10 mmHg or more, which according to the Mitral Valve Academic Research Consortium criteria defines iatrogenic LVOTO (10). In this context, when measuring for the Tendyne (Abbott Structural, Santa Clara, CA, USA) or Intrepid (Medtronic, Minneapolis, MN, USA) system, the minimized stent frame projection to the outflow tract would result in a slightly larger size NeoLVOT.

Surgical options for MAC-related mitral valve dysfunction are associated with operative mortality rates of 14% or higher (8). Although isolated MR could be treated with repair only, which is associated with a better prognosis and a lower rate of complications, these patients more commonly present mixed disease requiring a replacement approach (11). A range of resect and respect surgical options are available for mitral valve repair or replacement, with the former involving extensive *en bloc* resection of annular calcium, and the latter preferentially working around the calcium to avoid the complications associated with annular debridement (12). Alternative approaches like atrial-to-left ventricular valved conduits from the left atrium to the left ventricle for bypassing the mitral valve have also been described (13).

Our patient did not have an acceptable surgical risk, and the pre-procedural CT study moreover showed complete MAC, deeming her not eligible for conventional mitral valve surgery because of the increased risk for AV groove disruption. The heart team therefore opted for a transcatheter approach, choosing a Tendyne prosthesis.

In fact, percutaneous or hybrid approaches have been evaluated to try to minimize the operative risk. ViMAC approaches using a hybrid (transatrial or transapical) or a fully percutaneous strategy (transseptal ViMAC) are of interest in this scenario, but are not without their own challenges. As the anterior mitral leaflet (AML) is left *in situ*, both the transseptal and the transapical approaches are associated with a risk of LVOTO, particularly in patients with a smaller left ventricle, septal hypertrophy (14), longer or redundant anterior mitral valve leaflets, and a smaller AMA angle. In the TMVR registry, patients submitted to ViMAC TMVR had a lower overall technical success rate (62.1%) compared with valve-in-valve (94.4%) or valve-in-ring patients (80.9%), mainly because of LVOTO, which occurred in up to 39.7% of the patients. All-cause mortality was high at 30 days (34.5%) and 1 year (62.8%) (15). A meta-analysis of 13 studies involving ViMAC TMVR with balloon-expandable heart valves reported a median rate of LVOTO of 11.2%, a 3.7% incidence of transcatheter heart valve embolization, and a 4.1% incidence of moderate-to-severe PVL (16). Dedicated systems have been developed for TMVR, such as the transapical dual-stent frame Tendyne prosthesis (Abbott Structural, Santa Clara, CA, USA), which has been used in our report, with promising results according to a recent study (all-cause mortality rate of 5% and

40% at 30 days and 1 year, respectively, PVL and embolization rates of 0%, and an LVOTO rate of 5%) (17); moreover, LVOTO together with annular size are the mains reasons for screen failure with this device.

While not fully expected, in our clinical case TMVR resulted in LVOTO days after the procedure; the condition proved refractory to medical treatment and was clinically significant, resulting in decompensated HF. The position and angulation of the Tendyne toward LVOT is not always the same as it was predicted on the simulation, because the entry point can be more anterior than expected. Likewise, LV size can decrease after MR elimination and this fact can modify the relationship between the frame and the LVOT (18), and probably a combination of these two factors could influence the late development of LVOTO.

In patients with an increased risk of LVOTO, several techniques can be adopted preemptively or after its diagnosis to tackle both of the main underlying mechanisms: AML displacement and/or basal septal hypertrophy. For the former, intentional laceration of the AML to prevent LVOTO (the LAMPOON procedure) with an electrified guide wire can be an option (19). Another possibility would be balloon-assisted translocation of the mitral anterior leaflet (BATMAN). For basal septal hypertrophy, effective measures may comprise transcoronary ablation of septal hypertrophy (TASH) or the Septal Scoring Along the Midline Endocardium (SESAME) technique to perform an electrosurgical myotomy (20). In this case, ASA was successfully performed, achieving a reduction of the LVOT gradient of up to 16 mmHg. Different series report immediate improvements in gradient measurements following the procedure (21). There is a current shift in paradigm toward a more prophylactic approach to mitigate TMVR-induced LVOTO in high-risk patients (22), although this is not without pitfalls, because it increases the risk of permanent pacemaker implantation (from 16.7% to 35%) (23).

Mention should also be made of the possibility of using other TMVR devices with specific designs to prevent anterior leaflet displacement. Examples of this kind are supra-annular devices, such as the AltaValve System (4C Medical Technologies, Minneapolis, MN, USA), which could help mitigate the drawback of LVOTO, as it has a spherical nitinol frame sized to fit the left atrium, where it fixates, avoiding interaction with it. The first cases have been reported using a transapical approach, showing that it is a safe and effective device (24), and recently a small series of transseptal cases have been described, with excellent results (25).

Mitral annular calcification-related valve dysfunction is a high-risk condition associated with a poor prognosis and few simultaneously effective and safe therapeutic options. Appropriate pre-procedural risk evaluation by an interdisciplinary heart team and including multimodality imaging is mandatory to tailor therapeutic allocation to surgery, transcatheter techniques, or medical treatment only. Predicting potential hazards associated with each technique, such as LVOTO, allows the definition of preventive or bailout approaches such as TASH, LAMPOON, or SESAME. Other dedicated

devices with intra-atrial fixation have been developed, limiting the occurrence of this dreaded complication. A larger body of data is needed to define the best strategy in each case for this particularly challenging patient population.

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

BC-Q: Investigation, Resources, Writing – original draft, Writing – review & editing. CG: Investigation, Resources, Supervision, Validation, Visualization, Writing – original draft. JE-M: Conceptualization, Methodology, Writing – review & editing. RE-L: Supervision, Writing – original draft, Writing – review & editing. MB-P: Conceptualization, Methodology, Supervision, Writing – review & editing. RG-F: Conceptualization, Investigation, Writing – review & editing. FE-C: Supervision, Writing – review & editing. JL: Investigation, Supervision, Validation, Writing – review & editing. JB: Investigation, Supervision, Validation, Writing – review & editing. AÍ-R: Supervision, Validation, Writing – review & editing.

Funding

The authors 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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

Supplementary material

The Supplementary Material for this article can be found online at: <https://www.frontiersin.org/articles/10.3389/fcvm.2024.1431639/full#supplementary-material>

SUPPLEMENTARY FIGURE 1

Section A: TTE four-chamber view showing a double mitral lesion with MAC. Section B: TEE color comparison over mitral valve in mid esophageal view. Section C: 3D mitral *en face* view with and without color.

SUPPLEMENTARY FIGURE 2

CT analysis through 3Mensio software. (A) Annular segmentation in systole. (B) Smallest estimated LVOT. (C) MAC analysis and AMA angle. (D) Anterior mitral leaflet length.

SUPPLEMENTARY FIGURE 3

LVOT gradient evolution. Section A: TTE three-chamber LVOT gradient days after Tendyne implantation. Section B: TTE three-chamber LVOT gradient

after medical treatment before discharge. Section C: TTE three-chamber LVOT gradient 8 weeks after alcohol septal ablation.

SUPPLEMENTARY VIDEO 1

3Mensio Tendyne 29S LP simulation.

SUPPLEMENTARY VIDEO 2

Mitral valvuloplasty with Inoue balloon.

SUPPLEMENTARY VIDEO 3

29S LP Tendyne deployment, TEE commissural A2/P2 X plane view.

SUPPLEMENTARY VIDEO 4

LVOT and paravalvular leak evaluation after Tendyne deployment through TEE three-chamber view.

SUPPLEMENTARY VIDEO 5

TTE five-chamber view with 29SP LP Tendyne medial frame touching the septal bulge.

SUPPLEMENTARY VIDEO 6

Angiogram of the left anterior descending artery, showing second septal perforation artery.

References

1. Iung B, Delgado V, Rosenhek R, Price S, Prendergast B, Wendler O, et al. Contemporary presentation and management of valvular heart disease: the EUROSobservational research programme valvular heart disease II survey. *Circulation*. (2019) 140(14):1156–69. doi: 10.1161/CIRCULATIONAHA.119.041080
2. Van Hemelrijck M, Taramasso M, Gülmез G, Maisano F, Mestres CA. Mitral annular calcification: challenges and future perspectives. *Indian J Thorac Cardiovasc Surg.* (2020) 36(4):397–403. doi: 10.1007/s12055-019-00910-2
3. Abramowitz Y, Jilaihawi H, Chakravarty T, Mack MJ, Makkar RR. Mitral annulus calcification. *J Am Coll Cardiol.* (2015) 66(17):1934–41. doi: 10.1016/j.jacc.2015.08.872
4. Sorajja P, Gössl M, Babaliaros V, Rizik D, Conradi L, Bae R, et al. Novel transcatheter mitral valve prosthesis for patients with severe mitral annular calcification. *J Am Coll Cardiol.* (2019) 74(11):1431–40. doi: 10.1016/j.jacc.2019.07.069
5. Agrawal A, Reardon MJ, Goel SS. Transcatheter mitral valve replacement in patients with mitral annular calcification: a review. *Heart Int.* (2023) 17(1):19–26. doi: 10.17925/HI.2023.17.1.19
6. Guerrero M, Wang DD, Pursnani A, Eleid M, Khalique O, Urena M, et al. A cardiac computed tomography-based score to categorize mitral annular calcification severity and predict valve embolization. *JACC Cardiovasc Imaging*. (2020) 13(9):1945–57. doi: 10.1016/j.jcmg.2020.03.013
7. Demal TJ, Conradi L. Management of valve dysfunction in patients with mitral annular calcification. *Heart.* (2023) 109(21):1639–46. doi: 10.1136/heartjnl-2022-321572
8. Chehab O, Roberts-Thomson R, Bivona A, Gill H, Patterson T, Pursnani A, et al. Management of patients with severe mitral annular calcification: JACC state-of-the-art review. *J Am Coll Cardiol.* (2022) 80(7):722–38. doi: 10.1016/j.jacc.2022.06.009
9. Besola L, Falcetta G, Ceravolo G, Fiocco A, Colli A. New echocardiographic screening tool for left ventricular tract obstruction risk assessment in TMVR. *Int J Cardiol.* (2024) 408:132112. doi: 10.1016/j.ijcard.2024.132112
10. Reid A, Ben Zekry S, Turaga M, Tarazi S, Bax JJ, Wang DD, et al. Neo-LVOT and transcatheter mitral valve replacement: expert recommendations. *JACC Cardiovasc Imaging*. (2021) 14(4):854–66. doi: 10.1016/j.jcmg.2020.09.027
11. El-Eshmawi A, Alexis SL, Sengupta A, Pandis D, Rimsukcharoenchai C, Adams DH, et al. Surgical management of mitral annular calcification. *Curr Opin Cardiol.* (2020) 35(2):107–15. doi: 10.1097/HCO.0000000000000718
12. Said SM, Schaff HV. An alternate approach to valve replacement in patients with mitral stenosis and severely calcified annulus. *J Thorac Cardiovasc Surg.* (2014) 147(6):e76–8. doi: 10.1016/j.jtcvs.2014.02.039
13. Wright JS, Thomson DS, Warner G. Mitral valve bypass by valved conduit. *Ann Thorac Surg.* (1981) 32(3):294–6. doi: 10.1016/S0003-4975(10)61055-7
14. Elmariah S, Delaney JA, Bluemke DA, Budoff MJ, O'Brien KD, Fuster V, et al. Associations of LV hypertrophy with prevalent and incident valve calcification: multi-ethnic study of atherosclerosis. *JACC Cardiovasc Imaging*. (2012) 5(8):781–8. doi: 10.1016/j.jcmg.2011.12.025
15. Yoon SH, Whisenant BK, Bleiziffer S, Delgado V, Dhoble A, Schofer N, et al. Outcomes of transcatheter mitral valve replacement for degenerated bioprostheses, failed annuloplasty rings, and mitral annular calcification. *Eur Heart J.* (2019) 40(5):441–51. doi: 10.1093/eurheartj/ehy590
16. Alexis SL, Malik AH, El-Eshmawi A, George I, Sengupta A, Kodali SK, et al. Surgical and transcatheter mitral valve replacement in mitral annular calcification: a systematic review. *J Am Heart Assoc.* (2021) 10(7):e018514. doi: 10.1161/JAHA.120.018514
17. Gössl M, Thourani V, Babaliaros V, Conradi L, Chehab B, Dumonteil N, et al. Early outcomes of transcatheter mitral valve replacement with the Tendyne system in severe mitral annular calcification. *EuroIntervention.* (2022) 17(18):1523–31. doi: 10.4244/EIJ-D-21-00745
18. Guerrero ME, Grayburn P, Smith RL 2nd, Sorajja P, Wang DD, Ahmad Y, et al. Diagnosis, classification, and management strategies for mitral annular calcification: a heart valve collaborative position statement. *JACC Cardiovasc Interv.* (2023) 16(18):2195–210. doi: 10.1016/j.jcin.2023.06.044
19. Babaliaros VC, Greenbaum AB, Khan JM, Rogers T, Wang DD, Eng MH, et al. Intentional percutaneous laceration of the anterior mitral leaflet to prevent outflow obstruction during transcatheter mitral valve replacement: first-in-human experience. *JACC Cardiovasc Interv.* (2017) 10(8):798–809. doi: 10.1016/j.jcin.2017.01.035
20. Khan JM, Bruce CG, Greenbaum AB, Babaliaros VC, Jaimes AE, Schenke WH, et al. Transcatheter myotomy to relieve left ventricular outflow tract obstruction: the septal scoring along the midline endocardium procedure in animals. *Circ Cardiovasc Interv.* (2022) 15(6):e011686. doi: 10.1161/CIRCINTERVENTIONS.121.011686
21. Guerrero M, Wang DD, Hibert D, Urena M, Pursnani A, Kaddissi G, et al. Short-term results of alcohol septal ablation as a bail-out strategy to treat severe left ventricular outflow tract obstruction after transcatheter mitral valve replacement in patients with severe mitral annular calcification. *Catheter Cardiovasc Interv.* (2017) 90(7):1220–6. doi: 10.1002/ccd.26975
22. Wang DD, Guerrero M, Eng MH, Eleid MF, Meduri CU, Rajagopal V, et al. Alcohol septal ablation to prevent left ventricular outflow tract obstruction during transcatheter mitral valve replacement: first-in-man study. *JACC Cardiovasc Interv.* (2019) 12(13):1268–79. doi: 10.1016/j.jcin.2019.02.034
23. Elhadi M, Guerrero M, Collins JD, Rihal CS, Eleid MF. Safety and outcomes of alcohol septal ablation prior to transcatheter mitral valve replacement. *J Soc Cardiovasc Angio Interv.* (2022) 1(5):100396. doi: 10.1016/j.jscai.2022.100396
24. Goel SS, Zuck V, Christy J, Nallamothu N, Jagtap P, Gao J, et al. Transcatheter mitral valve therapy with novel supra-annular AltaValve: first experience in the United States. *JACC Case Rep.* (2019) 1(5):761–4. doi: 10.1016/j.jaccas.2019.10.034
25. Ninios V, Ninios I, Ranard LS, Vahl TP, Wróbel K. Transcatheter transseptal treatment of patients with severe mitral regurgitation using an atrial fixation mitral valve replacement technology. *Cardiovasc Revasc Med.* (2024) 58:25–30. doi: 10.1016/j.carrev.2023.07.006

Frontiers in Cardiovascular Medicine

Innovations and improvements in cardiovascular treatment and practice

Focuses on research that challenges the status quo of cardiovascular care, or facilitates the translation of advances into new therapies and diagnostic tools.

Discover the latest Research Topics

See more →

Frontiers

Avenue du Tribunal-Fédéral 34
1005 Lausanne, Switzerland
frontiersin.org

Contact us

+41 (0)21 510 17 00
frontiersin.org/about/contact



Frontiers in
Cardiovascular Medicine

