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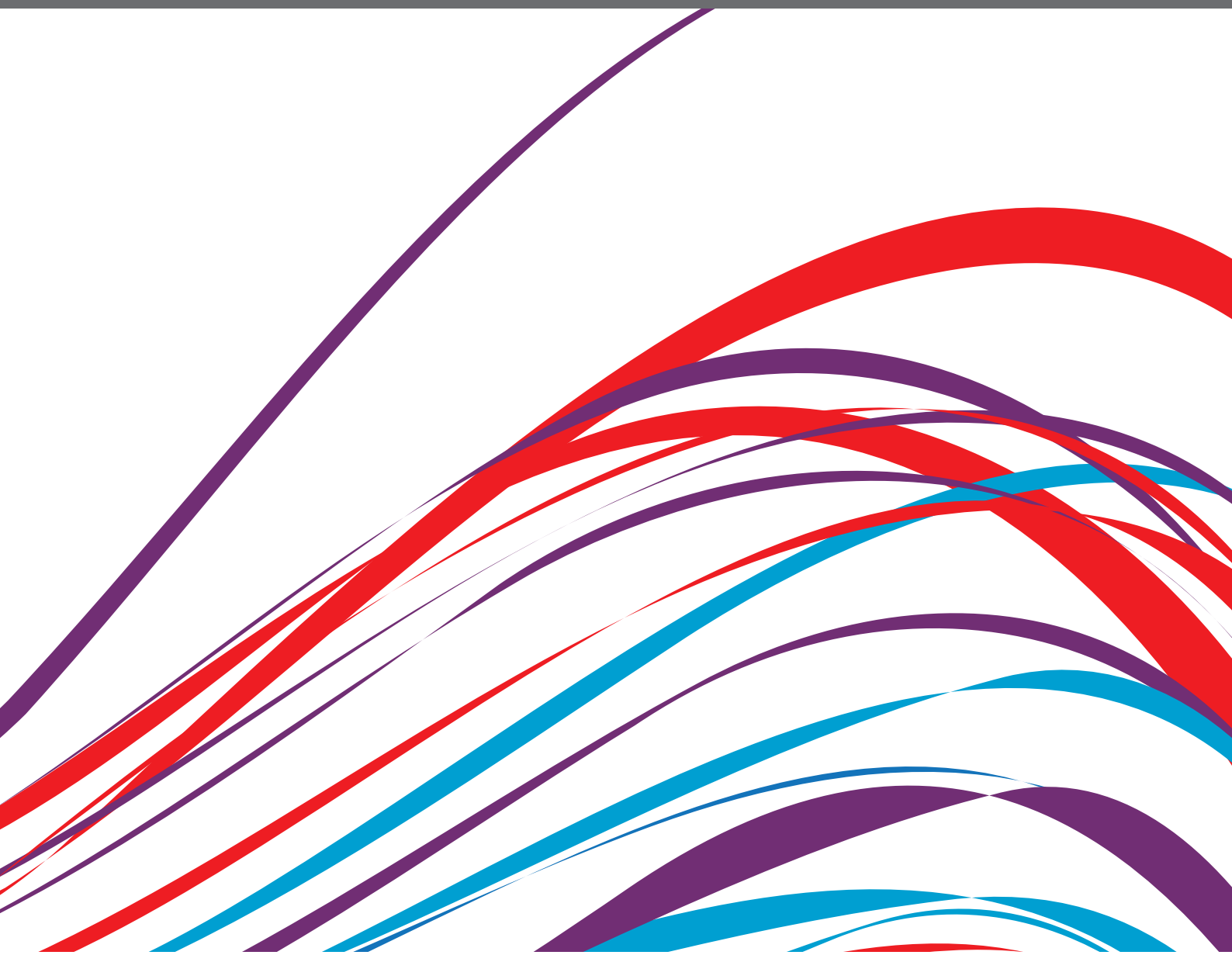

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CASE REPORTS IN HEART SURGERY: 2021

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CASE REPORT
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Case Report: Pericardial Effusion Treated With Pericardiectomy Plus Right Atrial Mass Resection: A 2-Year Follow-Up of Cardiac Rosai-Dorfman Disease

Edoardo Conte[1,2], *Antonio Brucato*[3], *Francesco Petrella*[4,5], *Emanuela Passoni*[6], *Gianfranco Lauri*[1], *Mauro Bigliardi*[7], *De Camilli Elisa*[4], *Gabriella Ricciardi*[1], *Carlo Selmi*[8,9], *Piergiuseppe Agostoni*[1,10], *Francesco Alamanni*[1,10†] *and Daniele Andreini*[1,10*†]

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Background: Rosai-Dorfman disease (RDD) is rare a sinus histiocytosis typically causing lymphadenopathy. Heart involvement is anecdotal, and <30 cases of cardiac RDD (cRDD) have been reported so far.

Case Presentation: A 46-year old woman with positive clinical history for RDD was admitted to our cardiology department with transthoracic echocardiography diagnosis of severe pericardial effusion and right atrial masses. Pericardiocentesis with catheter insertion was performed 3 days after the admission due to clinical evidence of cardiac tamponade. After 10 weeks of maximal medical therapy for inflammatory pericarditis, including non-steroidal anti-inflammatory drugs (NSAIDs), colchicine, steroids, and anakinra, at least 100 ml of pericardial citric liquid has been daily drained suggesting no clinical improvement. Pericardial liquid analysis demonstrated no malignant cells, but immunohistochemical analysis resulted positive for AE1–AE3, D2–40, S100, and CD68 consistent with an RDD diagnosis. Surgical management was judged clinically indicated, and 2 months after admission, the patient underwent pericardiectomy and debulking of atrial mass with freezing of remaining atrial neoformation. Regular clinical and echocardiography evaluation was performed without pericardial effusion recurrence after 2 years of follow-up.

Conclusions: This is the first case ever reported of cRDD who survived after 2 years of follow-up. Pericardiectomy could be feasible and effective for recurrent pericardial effusion in cRDD. Close follow-up and a multidisciplinary environment is needed to take care of cRDD patients.

Keywords: Rosai-Dorfman disease, pericardial effusion, pericardiectomy, case report, right atrial mass

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BACKGROUND

Rosai-Dorfman disease (RDD) is a rare clinical condition characterized by histiocyte proliferation causing lymphadenopathy, firstly described in 1969 (1). The cause of the disease is unknown, and diagnosis is based on specific immunohistochemical pattern with histiocytes that result to be S100 positive, CD68 positive, and CD1a negative; these characteristics are always present across all the wide spectrum of disease presentation. Previous reports suggest a potential association with HHV-6 and Epstein-Barr virus without further confirmation. Extra-nodal involvement is very rare, and cardiac involvement (cardiac RDD, cRDD) is anecdotal with <30 cases described so far, mostly in adult. Intra-cardiac masses and pericardial or epicardial involvement have been previously described, but the disease could arise from any cardiac site. Cardiac masses is the most common finding in patients with cRDD (2); among nine previous cases described, seven were treated with complete surgical excision, one with partial surgical excision and medical therapy with steroids, while in one patient, oral steroids was the sole conservative treatment. Of interest, previous reports included only two patients with recurrent pericardial effusion, similar to our patient; both patients died due to cRDD and diagnosis was autoptic. Most of previous reports had follow-up limited to few months after first diagnosis, while the only case treated conservatively showed the absence of mass volume increase after 2 years of follow-up. This case presented with both pericardial effusion and cardiac masses, and pericardiectomy plus cardiac masses partial excision was effectively performed for the first time ever in a patients affected by cRDD.

CASE REPORT

A 46-year old woman with positive clinical history for RDD with cutaneous involvement and arthritis, diagnosed 5 years before, was admitted to our cardiology department after transthoracic echocardiography diagnosis of severe pericardial effusion and right atrial masses (**Figure 1**). At admission, she referred dyspnea during ordinary activity (NYHA III), but no clinical signs of heart failure were present. Cardiac auscultation demonstrated soft heart sounds but pulsus paradoxus was detectable in the presence of sinus tachycardia and normal arterial pressure values (120/70 mmHg). Blood examination showed leukocytosis (WBC 25,000/μl, n.v. <10,000/ul) and increased inflammatory indexes (CRP 47.5 mg/l, n.v. <7 mg/l), while kidney and liver function indexes as well as BNP and Tn-I values were normal. Cardiac MRI confirmed the presence of severe pericardial effusion without concomitant signs of pericardial inflammation. Three separate right atrial neoformations were identified in the interatrial septum and near the right lateral atrioventricular junction (maximum size 13 × 19 mm involving the lateral wall of the right atrium). These masses appeared solid, with smooth borders, but with atrial wall infiltration; of note, superior cava vein involvement with caliber reduction was evident. Masses had inhomogeneous hyperintense signal at T2-weighted images with post-contrast enhancement (both first-pass perfusion images and late gadolinium enhancement). Overall, taking into consideration the positive medical history for RDD, cardiac MRI findings were consistent with cardiac involvement of RDD [(3, 4); **Figure 2**]. Possible differential diagnosis was primarily represented by cardiac metastasis from a primitive oncological disease, taking into consideration that this is the most common category of cardiac tumor, especially if multiple lesions are documented in the presence of pericardial effusion, like in this case. However, a total body positron emission tomography was performed and resulted negative for active oncological lesion.

Due to clinical evidence of cardiac tamponade 3 days after hospital admission, pericardiocentesis with catheter insertion was performed. Pericardial fluid analysis demonstrated no malignant cells, and immunohistochemical analysis showed the presence of histiocytes positive for AE1–AE3, D2–40, S100, and CD68 consistent with an RDD diagnosis.

After a total of 10 weeks of maximal medical therapy for inflammatory pericarditis including non-steroidal anti-inflammatory drugs (NSAIDs), colchicine, steroids, and anakinra according to current clinical guidelines, at least 100 ml of pericardial fluid has been daily drained suggesting no clinical improvement.

Surgical management of recurrent pericardial effusion was judged clinically indicated (5), and 2 months after admission, the patient underwent pericardiectomy and debulking of atrial mass with freezing of remaining atrial neoformation (**Figure 3**). In this scenario, although radical resection was not achievable, a debulking approach was indicated to prevent mediastinal syndrome due to complete SVC occlusion; this target, in fact, seemed to be more effectively obtained by surgery rather than radiotherapy. After successful resection, the portion of mass involving the superior caval vein was sent for histological analysis and resulted consistent with RDD (**Figures 3**, **4**). More precisely, the characteristic histiocytes were found with abundant and vacuolated cytoplasm, rounded nuclei with coarse chromatin, and a single prominent nucleolus. Emperipolesis was demonstrated as well (**Figure 5**). The post-operative course was uneventful, and the patient was dismissed 2 weeks after surgery in good clinical condition. Regular clinical and echocardiography evaluation was performed, and patient had no pericardial effusion. At follow-up, cardiac MRI confirmed the absence of pericardial effusion, the mild and slow progression of atrial masses in the presence of moderate-to-severe mitral regurgitation, and mild-to-moderate aortic regurgitation that was not present at the time of surgery and is possibly related to heart valve involvement of cRDD (**Figure 2**).

DISCUSSION AND CONCLUSION

Rosai-Dorfman is a rare disease causing massive lymphadenopathy due to histiocyte proliferation. Cardiac involvement is anecdotal with <30 cases described. Heart involvement has been previously described as an intra-cardiac mass, pericardial or epicardial involvement, and pulmonary arterial/aorta infiltration. Previous reports included only

Abbreviations: RDD, Rosai-Dorfman disease; NSAIDs, non-steroidal anti-inflammatory drugs; WBC, white blood cells; CRP, C-reactive protein; cRDD, cardiac RDD.

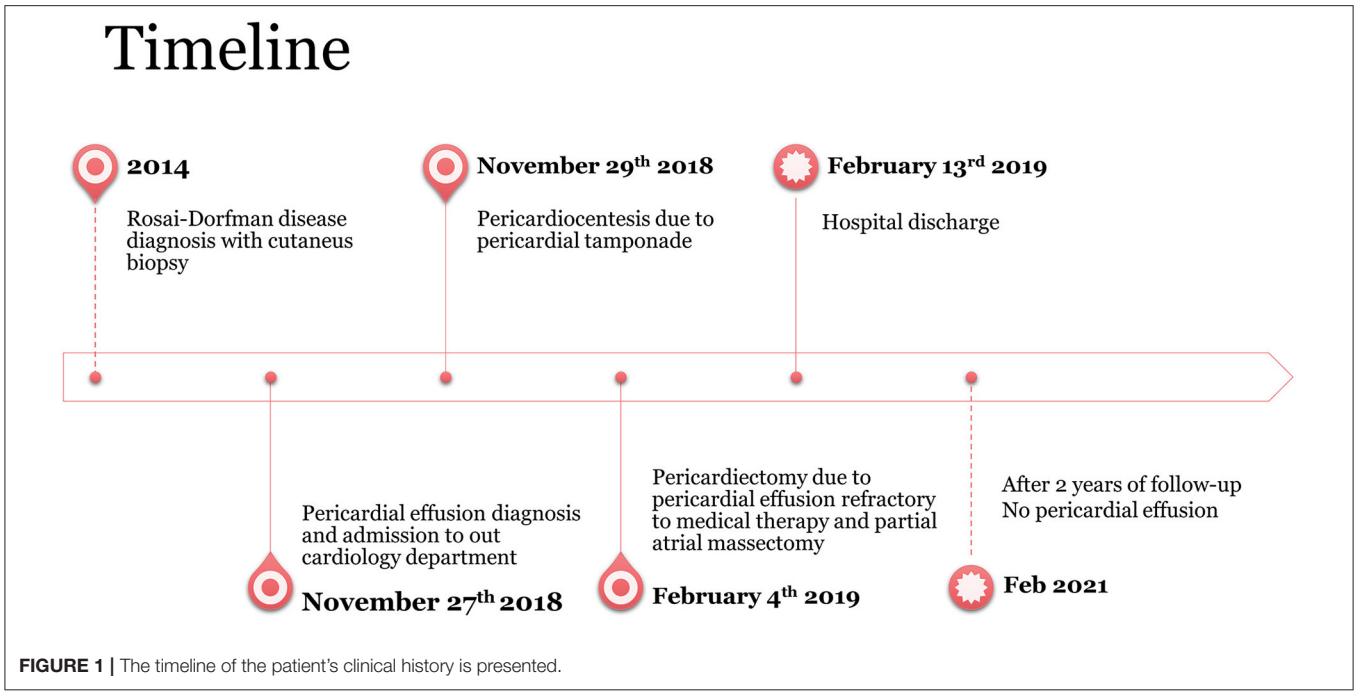

FIGURE 1 | The timeline of the patient's clinical history is presented.

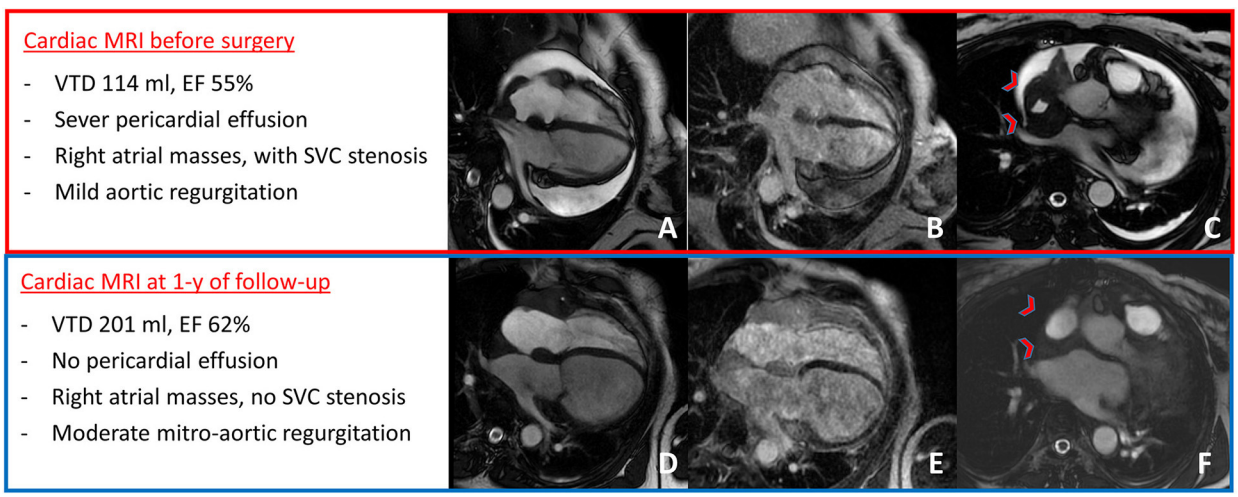

FIGURE 2 | Cardiac MRI at presentation showed severe pericardial effusion with mild aortic regurgitation **(A)**, right atrial masses with LGE **(B)**, and mass infiltration of the superior cava vein causing stenosis [red arrow-heads in **(C)**]. After 1 year of follow-up, cardiac MRI showed no pericardial effusion with moderate mitro-aortic regurgitation **(D)** and mild increase in right atrial masses' size **(E)**, but with no superior cava vein stenosis [red arrow-heads in **(F)**].

six patients with pericardial involvement of which three patients had recurrent pericardial effusion; two of them due to cRDD complications (2), while in the third report of patients presenting with pericardial tamponade, no clear information regarding prognosis is provided (6). Of interest, in a previously described case (7) presenting with both pleural and pericardial recurrent effusion, multiple pleural biopsies resulted negative for pathological findings, highlighting how the disease could be a diagnostic challenge even if suspected. Recently, another case of cRDD pericardial presentation (fibrinous pericarditis) has been described after autoptic evaluation, underlining the dismal prognosis of pericardial involvement from the disease, especially when unrecognized (8).

When cardiac masses are present, a complete excision has been previously described (9), but in our patient, it was not feasible due to the extensive atrial wall infiltration involving the tricuspid annulus as well. The present case is the first in which surgery was indicated for intractable recurrent pericardial effusion effectively treated with pericardiectomy; partial removal of the mass was performed as well, in order to avoid the post-surgical progression of superior caval vein stenosis. After 2 years of follow-up, the longest ever reported for cRDD, no pericardial effusion is detectable, confirming that pericardiectomy could be effective in cRDD, but the rapid progression of both mitral and aortic regurgitation from mild to moderate-to-severe entity, possibly due to RDD involvement,

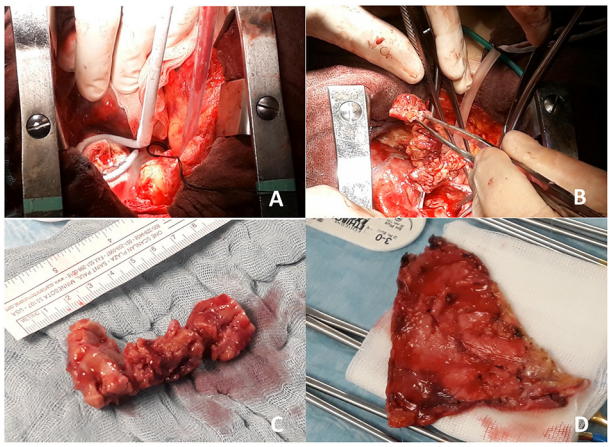

FIGURE 3 | Intraoperative view of residual mass freezing (**A** and **Supplementary Video 1**) and removal of superior cava vein mass (**B** and **Supplementary Video 2**). In **(C)**, gross surgical pathology of the removed mass (7 cm length) is shown, while in **(D)**, infiltrated pericardium is shown after pericardiectomy.

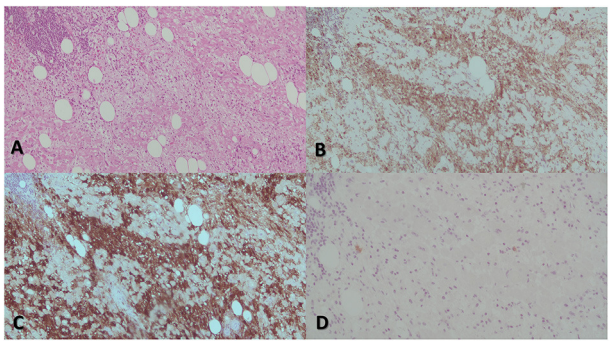

FIGURE 4 | The most prominent histological feature is expansion of the cardiac muscular cells that are filled with histiocytes **(A)**. These characteristically have abundant, often vacuolated cytoplasm, rounded nuclei with coarse chromatin, and a single prominent nucleolus. The atypical histiocytes express CD68 **(B)**. In contrast to reactive sinus histiocytes, they are S100 positive **(C)** but negative for CD1a **(D)**. All these features are consistent with RDD.

pointed out the aggressive nature of the disease itself and the need for close clinical and echocardiographic follow-up in this kind of patients.

In conclusion, even taking into consideration the limited possibility to extrapolate definite therapeutic strategies from a single case report, the main teaching points of the present case are that pericardiectomy could be feasible and effective in patients with RDD with cardiac involvement presenting with recurrent pericardial effusion; on the contrary, the progression of both mitral and aortic regurgitation pointed out the aggressive nature of the disease itself that needs a multidisciplinary evaluation and strict follow-up. Finally, as previously described, heart masses due to RDD seem to have a slow progression rate.

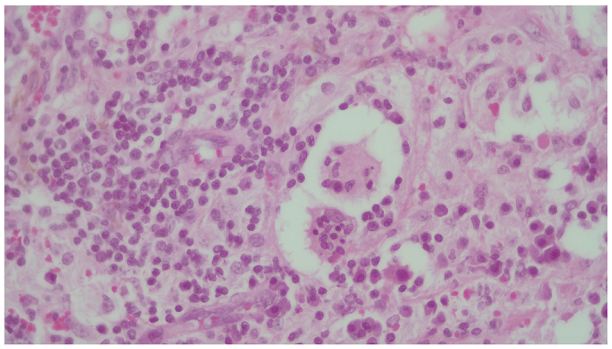

FIGURE 5 | The identification of emperipolesis, defined as the presence of an intact cell within the cytoplasm of another cell. This finding is reported to be typical of RDD and provides a differential diagnosis with PS100+ Erdheim-Chester disease.

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

Ethical review and approval was not required for the study on human participants in accordance with the local legislation and institutional requirements. The patients/participants provided their written informed consent to participate in this study. Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

AUTHOR CONTRIBUTIONS

EC and DA designed and wrote the first draft of the manuscript. AB, FP, PA, FA, and CS provided senior supervision and critically revised the design and conception of the manuscript. EP, GL, MB, GR, and DE provided important contribution in data collection, data analysis and interpretation. All authors critically revised the manuscript for important intellectual content, approved the final version of the manuscript and agreed to be accountable for all aspect of the work. All authors have a fundamental role in the clinical management of the case report.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fcvm.2021.668031/full#supplementary-material

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CASE REPORT
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Case Report: The Cox-Maze IV Procedure in the Mirror: The Use of Three-Dimensional Printing for Pre-operative Planning in a Patient With Situs Inversus Dextrocardia

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The safety and efficacy of the Cox-Maze IV procedure (CMP-IV) for situs inversus dextrocardia patients with atrial fibrillation is yet to be determined. Herein, we present the case of a 39-year-old male patient admitted to our cardiac center following progressive exertional dyspnea. The patient was diagnosed with situs inversus dextrocardia, severe mitral regurgitation, and paroxysmal atrial fibrillation. A three-dimensional (3D) heart model printing device embedded with designated ablation lines was used for pre-operative planning. Mitral valvuloplasty, CMP-IV, and tricuspid annuloplasty were performed. The patient had an uneventful recovery and was in sinus rhythm during a 12-month follow-up period using a 24-h Holter monitoring device. The case herein is one of the first to report on adopting the CMP-IV procedure for situs inversus dextrocardia patients with complex valvuloplasty operation. In addition, the 3D printing technique enabled us to practice the Cox-maze IV procedure, given the patient's unique cardiac anatomy.

Keywords: Cox-maze IV procedure, mirror-image dextrocardia, three-dimensional printing, pre-operative planning, surgery

BACKGROUND

The Cox-Maze IV procedure (CMP-IV) is the only operation and technology with an FDA-approved indication for the surgical treatment of AF. However, the safety and efficacy of the CMP-IV for situs inversus dextrocardia patients with atrial fibrillation is yet to be determined. The case herein is one of the first to report on adopting the CMP-IV procedure for mirror-image dextrocardia patients with complex valvuloplasty operation with the guidance of the 3D printing technique.

CASE PRESENTATION

A 39-year-old man who complained of progressive exertional dyspnea and intermittent palpitation was referred to our department. He denied any familiar history of situs inversus dextrocardia or other cardiac health comorbidities. The patient, however, had occasional dizziness but denied any history of hypertension or cerebral infarction. Physical examination revealed a systolic

blowing murmur (grade 3/6) at the fifth intercostal space lateral to the right midclavicular line. Chest roentgenogram and computed tomography (CT) showed dextrocardia with an enlarged silhouette and situs inversus totalis (**Figures 1A,B**). The anatomic diagnosis was mirror-image dextrocardia, L-loop ventricles, and typical related great arteries without associated congenital cardiac abnormalities (**Figures 1C,D**). Transesophageal echocardiography further revealed severe mitral valve regurgitation where the vena contracta was >0.7 cm and tricuspid regurgitation following annular dilation. The left atrium diameter was 5.8 cm without thrombosis formation (**Figures 1E,F** and **Supplementary Video 1**). A 24-h Holter monitoring demonstrated paroxysmal AF with a total burden of 165 min.

Three-dimensional (3D) printing of the heart was performed using the derived cardiac CT data to precisely comprehend the anatomy and guide surgical ablations. A stereolithography file of the 3D model embedded with designated ablation lines was generated (**Figure 2A**), which was then manufactured with soft and flexible resinous material at a ratio of 1:1 (**Figure 2B**). The easy-to-cut and retractable feature of the 3D model (**Supplementary Video 2**) allowed us to rehearse the CMP-IV pre-operatively (**Figure 2C**).

The cardiopulmonary bypass for the patient was routinely performed via a median sternotomy (**Figure 3A**). The operator operated on the patient's left side while the cardiopulmonary bypass machine was placed on the right side. A biatrial CMP-IV procedure with mitral valvuloplasty and tricuspid annuloplasty were simultaneously performed (**Supplementary Video 3**). The left atrium was accessed via the interatrial groove. We lengthened the mitral isthmus lesion to the posterior mitral valve annulus for the left atrial lesion sets and ablated the coronary sinus endocardially and epicardially with a bipolar radiofrequency pen (AtriCure Inc., Cincinnati, OH). Other lesions, which were created using bipolar radiofrequency clamps (AtriCure Inc., Cincinnati, OH), included: (i) bilateral pulmonary veins

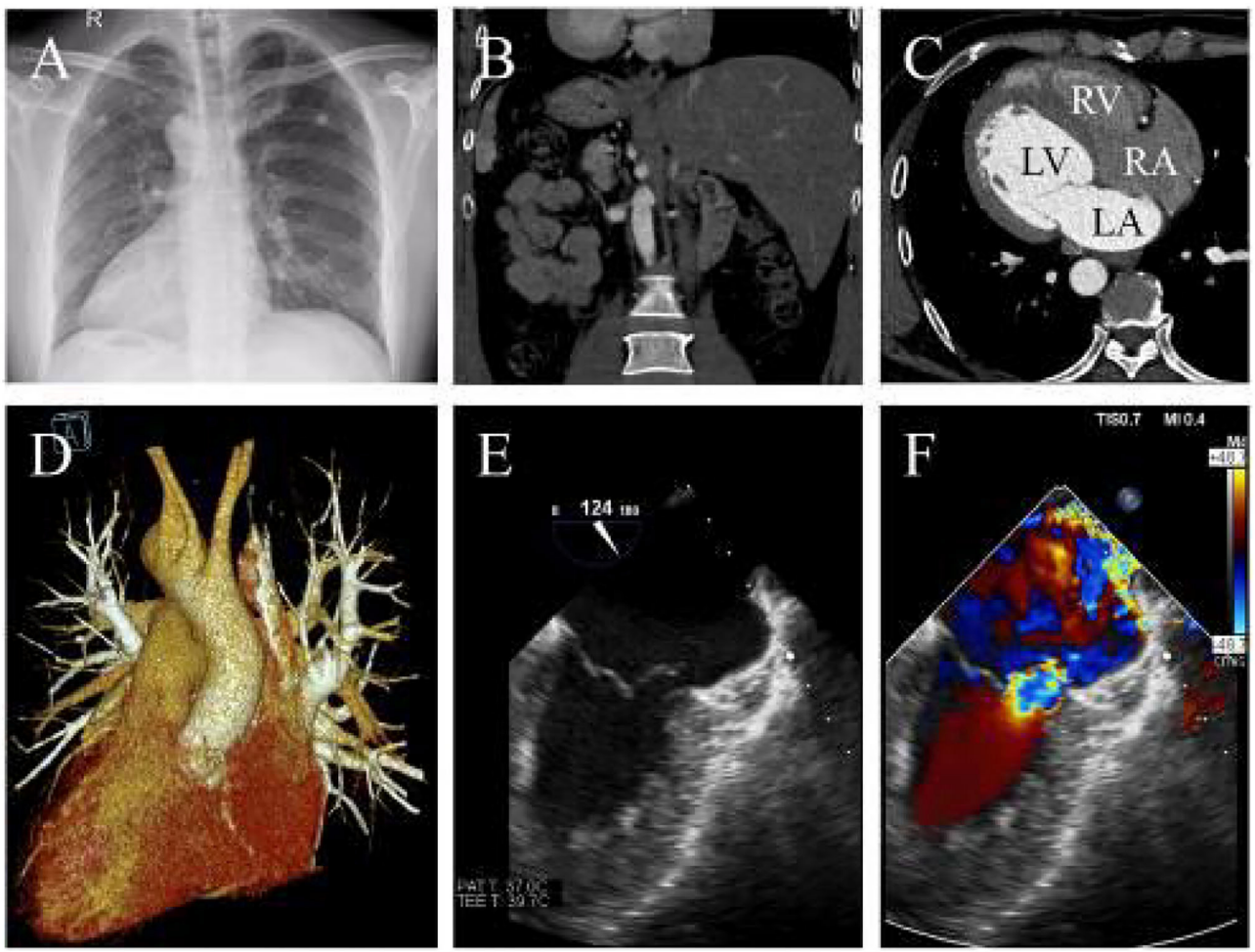

FIGURE 1 | (A) X-ray indicates dextrocardia and cardiac enlargement. **(B)** CTA reveals transposition of the abdominal organs and **(C)** confirming L-loop ventricular orientation and concordant atrioventricular connections. **(D)** Three-dimensional CTA shows normal great artery development and connections. **(E)** The transesophageal echocardiography shows mitral chordae rupture and **(F)** severe mitral regurgitation (LA, left atrium; LV, left ventricle; RA, right atrium; RV, right ventricle).

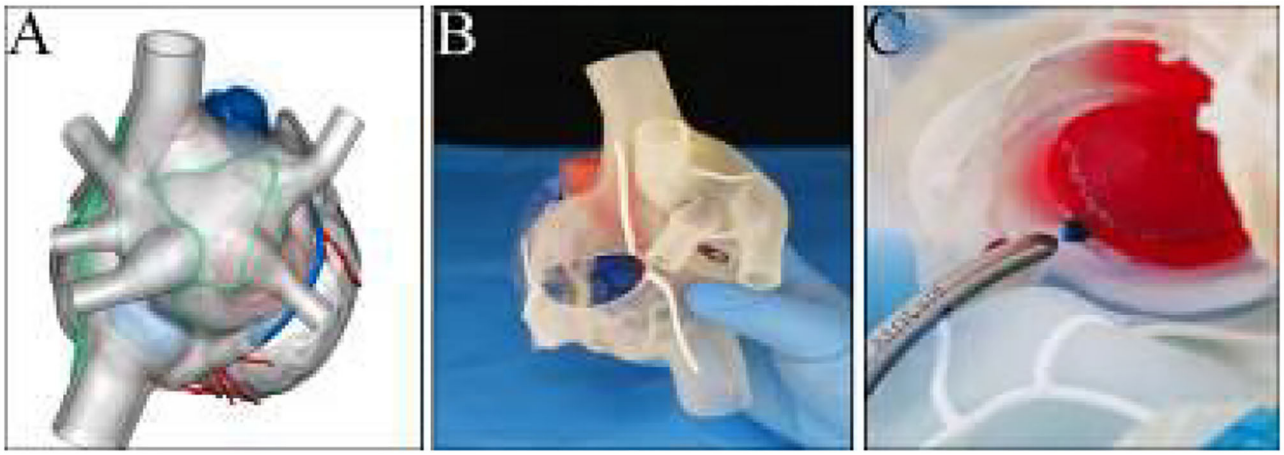

FIGURE 2 | (A) Pre-printing digital file of the 3D model embedded with designated ablation lines. **(B)** View of the three-dimensional printed model. **(C)** Clamping on the model of the mitral isthmus line during the rehearsal process.

isolation; (ii) ablation lines connecting the left atrial appendage and the left superior pulmonary vein; (iii) ablation lines connecting the right and the left superior pulmonary veins; (iv) ablation lines connecting the right and the left inferior pulmonary veins, and (v) mitral line lesions (**Figure 3B**). Finally, the ligament of Marshall was dissected, and the left atrial appendage was isolated using an epicardial AtriClip closure device. The entire right atrial lesion sets were created using bipolar radiofrequency clamps, including the annular tricuspid lesions (**Figure 3C**), superior and inferior vena cava lesion lines, and lesion lines connecting the right atrium incision to the right atrial appendage.

We performed A1 ruptured chordae resection, followed by implantation of a single flexible artificial chord with 4-0 Gore-tex *in situ*. The residual leak of the anterior commissure and the A2 leaflet cleft was closed accordingly. A 32 mm rigid ring (Element Force ARM32, Kingstron Bio, Suzhou, China) was then implanted to stabilize the mitral annulus. The coaptation height was 9 mm after reconstruction of the mitral valve (**Figure 3D**). After the closure of the interatrial groove incision and de-airing, the aortic clamp was removed, and the right atrium was entered via a longitudinal incision.

The tricuspid repair was obtained with a 28 mm band (SOVERING TRICUSPID BAND^{TM}, Sorin Group Italia Srl, Italy) due to the mirror-imaged anatomy. The patient was in sinus rhythm before cardiopulmonary bypass weaning, and no atrioventricular block was detected. He had an uneventful recovery, and the anti-arrhythmia and oral anticoagulation drugs were continued for 3-months post-operatively. The patient was in sinus rhythm (24-h Holter monitoring device) with an improved quality of life during a 12-month follow-up period.

DISCUSSION AND CONCLUSIONS

Mirror-image dextrocardia is characterized by a mirror-image change of the normal heart mostly accompanied by situs inversus viscerum with only 3–10% intracardiac anomaly (1). The CMP-IV procedure is the only operation and technology with an FDA-approved indication for the surgical treatment of AF. The term "Maze" is appropriately used only to refer to the biatrial lesion sets of the CMP-IV. Less extensive lesion sets should not be referred to as the CMP-IV but rather as a surgical ablation (2).

Studies have confirmed that the Cox-maze IV procedure concomitant with valvular surgery had higher efficiency concerning AF elimination, AF burden relief, stroke rate control, long term survival rate, and health-related quality of life improvement than valvular surgery alone, catheter ablation, and other forms of surgical AF ablation (3–5). CMP-IV showed excellent efficacy in restoring sinus rhythm during the 10-years follow-up in the studies mentioned above. The procedure was performed in patients with paroxysmal and non-paroxysmal AF (6). However, age, AF duration, non-paroxysmal AF, and left atrial size were adverse prognostic factors (6, 7) for the long-term outcome of the Cox-Maze IV procedure. Surgical ablation experiences for AF patients with dextrocardia have been sporadically reported (8). Due to the low incidence rate, there is a paucity of data describing the safety and efficacy of CMP-IV *in situs* inversus dextrocardia patients, let alone its long-term outcomes and durability.

The ablation device used herein was the bipolar radiofrequency clamps combined with a bipolar radiofrequency pen, given cryothermal energy is not available in China. We performed bipolar radiofrequency clamping in three sets for each lesion line with two applications per set without unclamping, which has been documented to result in 100% atrial lesion transmurality (9). A complete set of right atrial lines was performed with bipolar radiofrequency (10). Dissection of the epicardial fat of the right atrioventricular groove not only warranted tricuspid annular clamping but also avoided right coronary artery injury. Incomplete ablation of mitral isthmus had been reported to cause post-operative atrial flutters (11). The painting and stamping ablation technique was used for mitral

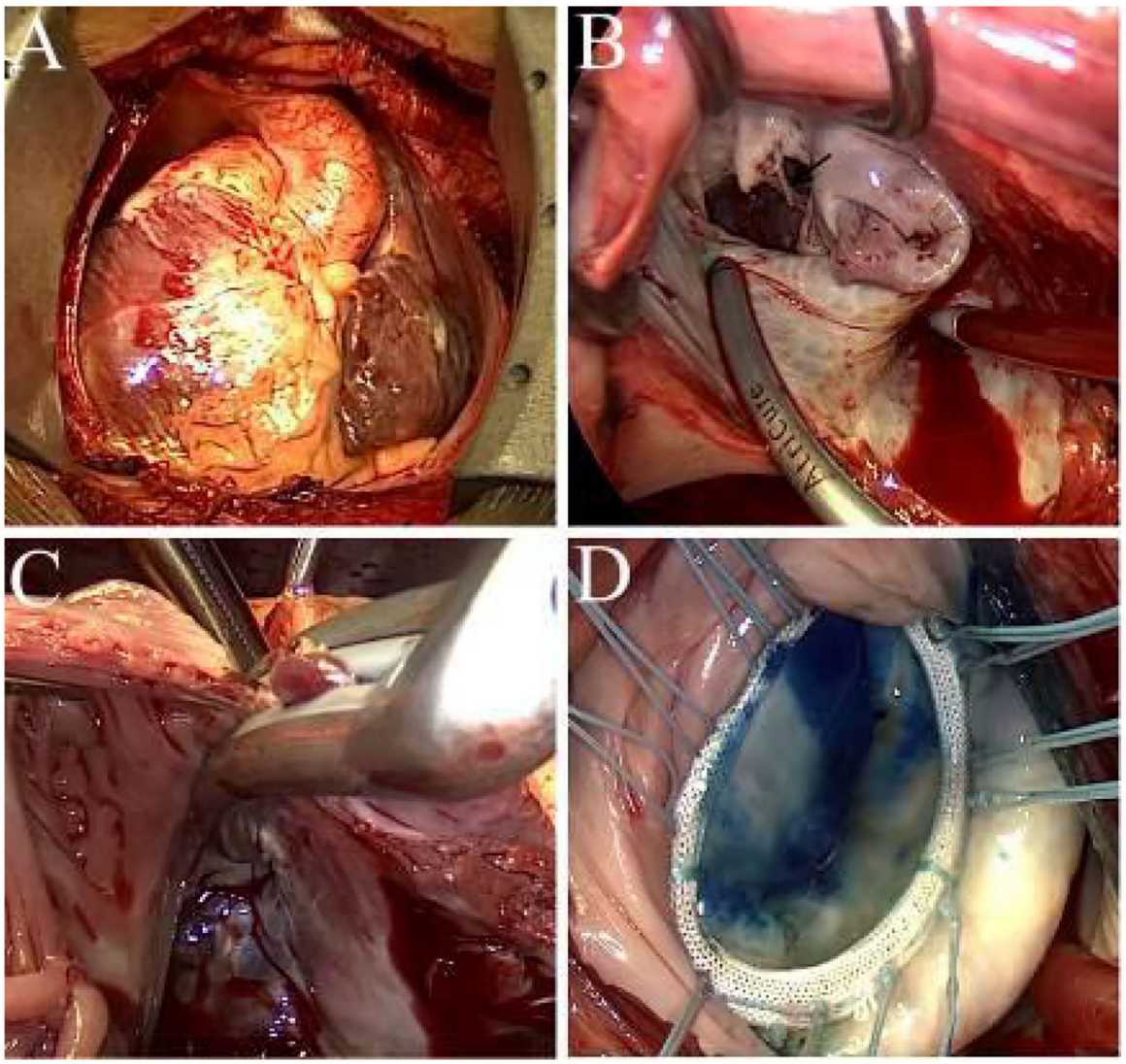

FIGURE 3 | (A) Intraoperative view of the mirror-image dextrocardia. **(B)** Ablation at the mitral line, black arrow indicates the ruptured chordae. **(C)** Ablation to the tricuspid annulus, the jaw of the bipolar clamps are placed across the tricuspid annulus. **(D)** Successful result of saline injection test after mitral valvuloplasty.

isthmus lesions with a bipolar radiofrequency pen given the location. No post-operative atypical atrial flutter was detected.

The pulmonary veins are a crucial source of ectopic beats, triggering frequent paroxysmal AF (12). However, the duration of AF directly influenced the degree of atrial remodeling and the complexity of the atrial substrate (13). Though the patient had paroxysmal AF, we decided to apply the CMP-IV procedure rather than the modified left atrial lesion set or a more straightforward pulmonary vein isolation. The decision was based on the fact that patients with extended paroxysmal AF may have an advanced underlying disease than realized, especially if there are signs of left atrial enlargement (14).

Currently, there is a knowledge-practice gap between Class-1 recommendation on concomitant surgical ablation for AF (2) and its low adoption rate (15). One of the barriers impeding the practice was the unfamiliar cardiac anatomy, which made the procedure challenging, extending the cardiopulmonary bypass and aortic cross-clamp time. The other stumbling block was that no standardized curriculum exists for training cardiothoracic surgery residents in surgical ablation for AF.

Exposure to complete ablations and familiarization with the anatomic boundaries may be accomplished in the way of simulation, tissue labs, or higher-fidelity models (16). In the case herein, the 3D printing model enabled a dynamic visualization of the spatial relationship between the specific ablation lines and the critical anatomic references such as the coronary sinus, the mitral isthmus, the posterior mitral annulus, and the right coronary artery. In addition, the rehearsal process provided an intuitive perspective concerning viable ablation strategies, including the location, direction, and length for each clamping

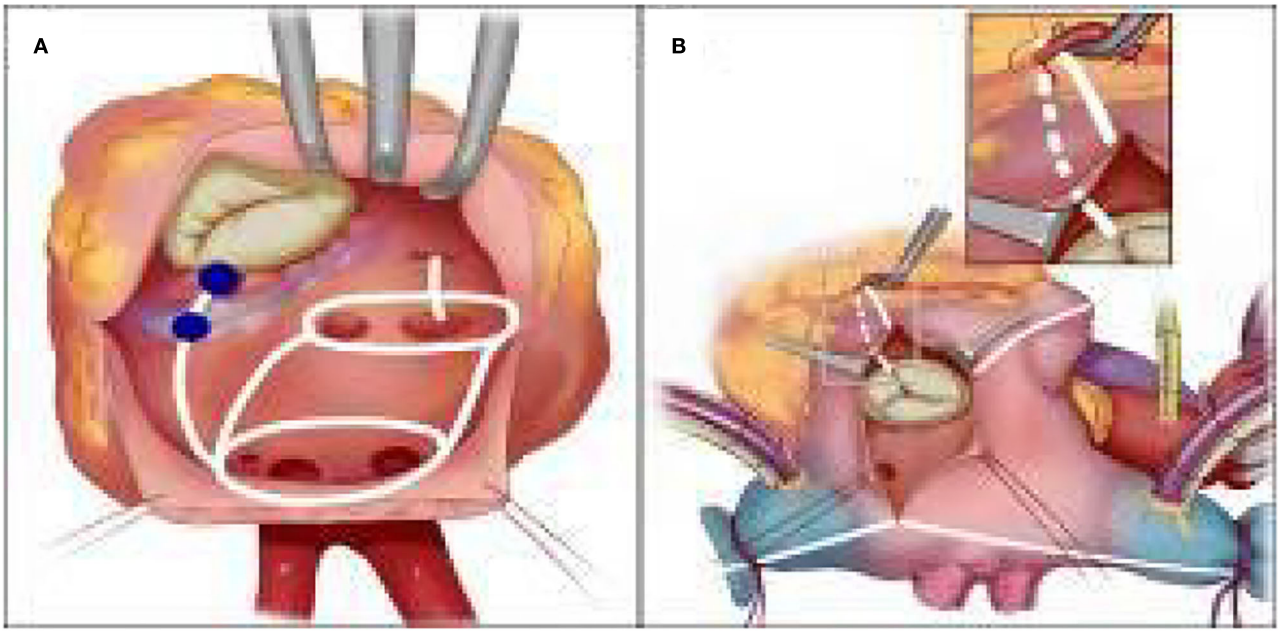

FIGURE 4 | (A) Left atrial lesion sets for Cox-maze IV procedure in the mirror-image dextrocardia, Mitral isthmus lesion is between the shaded color (light and dark blue) at the posterior mitral annulus and the coronary sinus. **(B)** Right atrial lesion sets for Cox-maze IV procedure in the mirror-image dextrocardia. The tricuspid annulus lesion is completed after dissecting the peripheral epicardial fat of the right coronary artery.

to ensure continuous lesion sets and avoid potential damages to the surrounding structures.

The Cox-Maze IV procedure in such a rare malformation was complex and technically challenging. We believe that dedicated pre-operative ablative lesion sets (**Figure 4**) and procedural rehearsal with 3D printing heart models are of added value given that it shortens operation time and increases the chance to achieve better results than the empirical "track in mind" does. Our individualized 3D printing model embedded with designated lesion lines may also serve as a potential candidate for clinical instruction in AF surgery.

Nonetheless, the study herein is without its limitations. The study is a single-patient case report, given the low incidence rate and the rarity of the disease. Our 3D printing heart model could not be used for a pre-operative mitral or tricuspid rehearsal valve repair. After the procedure, the lack of electroanatomical mapping is the only test to verify if the ablation lines were adequately performed and the desired block reached. However, the latest guidelines (2, 17) do not recommend routine intraoperative mapping for each lesion line of the Cox-maze IV procedure to prove if the derived electrophysiology was effective. We thus strongly believe that an in-depth collaboration between the cardiac surgeon and the electrophysiologist would widen the spectrum for the effective treatment of valvular AF in the future.

To our knowledge, the study herein is one of the first to adopt the CMP-IV procedure in a patient with situs inversus dextrocardia, which was successfully guided using rapid-prototyping techniques for the preparation of individualized lesion sets (18). In addition, the 3D printing model helps surgeons mimic and modify the CMP-IV procedure in patients with unfamiliar cardiac anatomy, particularly with rare malformations, supporting pre-operative planning and training (19) in surgical ablation for AF.

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 human participants were reviewed and approved by the Ethics Committee of the Second Xiangya Hospital of Central South University. The patients/participants provided their written informed consent to participate in this study. Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

AUTHOR CONTRIBUTIONS

LS and CF drafted the manuscript. LL designed the study. HZ, HL, CI, CL, and CF revised the manuscript. LS, HZ, and HL were

responsible for the collection of data or analysis. All authors read and approved the final manuscript.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fcvm.2021.722413/full#supplementary-material

Supplementary Video 1 | Transesophageal echocardiography showed severe mitral regurgitation.

Supplementary Video 2 | Demonstration of the material used for 3D printing.

Supplementary Video 3 | Descriptions of the complete lesion sets and critical steps for valvuloplasty.

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CASE REPORT
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Case Report: Intraoperative Open-Heart Coronary Angiography in Acute Type A Aortic Dissection

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For patients with acute type A aortic dissection, strongly suspected of having concomitant severe coronary artery disease (CAD), preoperative or intraoperative coronary angiography has been recommended. However, conventional selective coronary angiography in this setting may extend the dissection or aortic rupture. We present the use of intraoperative open-heart coronary angiography in a patient with acute type A aortic dissection. A 50-year-old man presented with chest pain and dyspnea and was admitted to our department with acute type A aortic dissection. The patient underwent coronary artery stent implantation in the left anterior descending coronary artery (LAD) 3 years previously due to an acute myocardial infarction. This time we failed to evaluate the patency of the LAD using multidetector computed tomography. An aortic rupture occurred due to conventional coronary angiography, and open-heart coronary angiography was performed. The examination revealed no significant stenosis. A Bentall procedure and total aortic arch replacement were performed, with an intraoperative stent inserted into the descending aorta, and the patient had an uneventful postoperative course. From this case, we learn that intraoperative open-heart coronary angiography is safe and effective in patients with acute type A aortic dissection.

Keywords: open-heart, coronary angiography, coronary artery disease, aortic dissection, case report

INTRODUCTION

Acute type A aortic dissection is a life-threatening condition associated with high mortality rates (1). Patients with this disease usually have the same risk factors for coronary artery disease (CAD), and a study has shown that 25% of patients with acute type A aortic dissection have evidence of severe coronary atherosclerosis (2). In cases of acute type A aortic dissection, CAD remains a major predictive factor for both early and late mortality (3, 4) and if possible, it should be identified accurately. Although coronary computed tomography angiography may reveal the conditions of the coronary arteries in most cases, coronary angiography remains the criterion standard in the evaluation of CAD (3, 4). For selected patients with acute type A aortic dissection, strongly

suspected of having concomitant severe CAD, preoperative or intraoperative coronary angiography has been recommended by some authors (5–7).

However, conventional selective coronary angiography in this setting may extend the dissection or aortic rupture by advancing the catheter into the false lumen (8). Therefore, a special type of coronary angiography should be performed in these cases. Intraoperative open-heart coronary angiography has been described in a canine model and one patient with aortic valve endocarditis (9, 10). This article will present the use of it in a patient with acute type A aortic dissection and this may be the first time the open-heart coronary angiography was used in this disease.

CASE

A 50-year-old man presented with chest pain and dyspnea and was admitted to our department with acute type A aortic dissection. The patient underwent coronary artery stent implantation in the left anterior descending coronary artery (LAD) 3 years previously, to treat an acute myocardial infarction. Multidetector computed tomography (MDCT) revealed a severe dissection of the aortic sinus, but evaluating the patency of the LAD was made difficult because of interference from the coronary stent (**Figures 1A,B**). Therefore, conventional coronary angiography was performed in our hybrid operating room in case of aortic rupture.

First, we established a cardiopulmonary bypass through the right axillary artery and vena cava. Second, a 5F JL4 angiographic catheter was threaded through the left femoral artery into the ascending aorta. However, an aortic rupture occurred when we tried to select the left coronary artery,

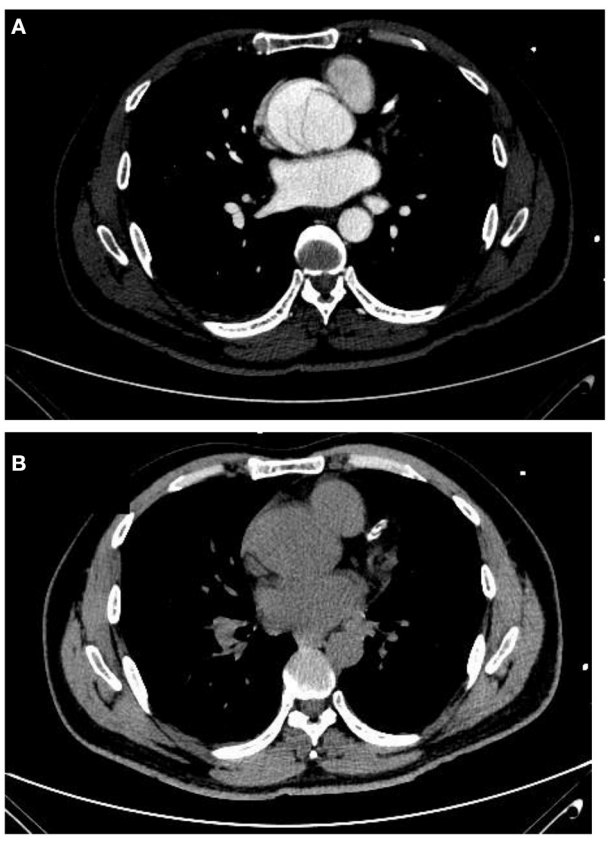

FIGURE 1 | (A) MDCT image of the LAD in an enhancement scan. **(B)** MDCT image of the LAD in a plain scan. LAD, left anterior descending coronary artery; MDCT, multidetector computed tomography.

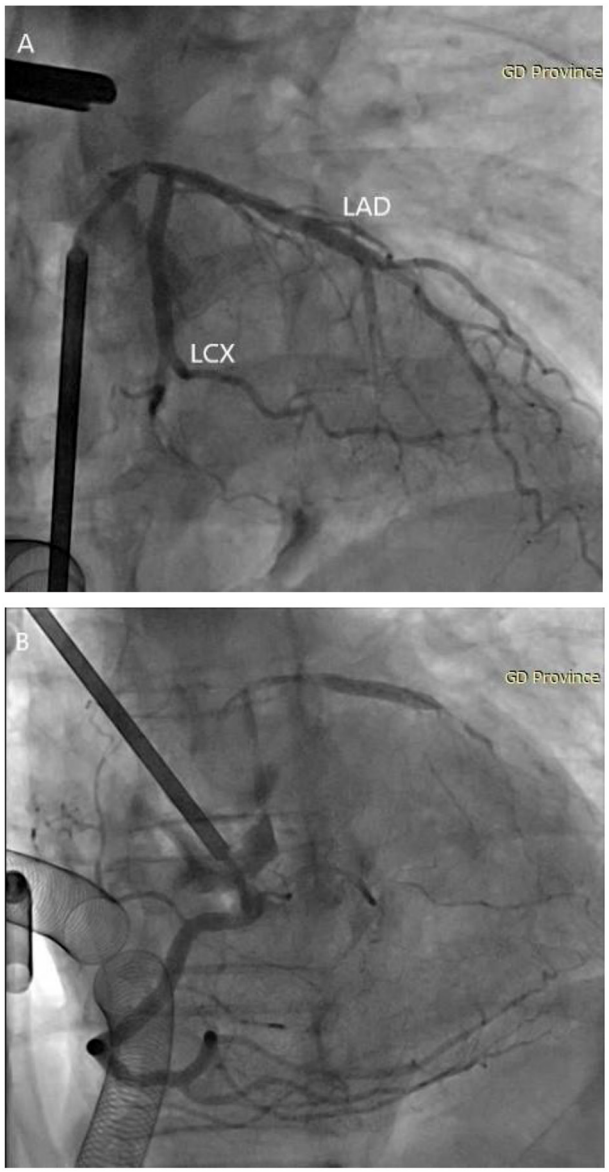

FIGURE 2 | Visualization of the coronary angiography. **(A)** Intraoperative open-heart coronary angiography shows no significant stenosis in the left coronary artery. **(B)** Intraoperative open-heart coronary angiography shows no significant stenosis in the right coronary artery. LAD, left anterior descending coronary artery; LCX, left circumflex coronary artery.

therefore an aortic cross-clamp was immediately performed. After perfusion of the cardioplegic solution (1,200 ml cold blood cardioplegia), we performed a coronary angiography by cannulation of both coronary ostia with selective cardioplegia catheters (12 and 14 Ch, Coronary Ostial Perfusion Cannula, Medtronic, Minneapolis, MN, USA). A pump was used to inject 5 milliliters of contrast medium (loperamide, 300 mg iodine/mL, Ultravist, Bayer, Berlin, Germany) at a pressure of 1,000 psi and a flow rate of 4 mL/s, showing no significant coronary artery stenosis in the coronary artery (**Figures 2A,B**). After injection of the contrast medium, the cardioplegic solution (1,200 ml) was infused through the coronary arteries to prevent contrast toxicity and cardioplegic washout.

Finally, the patient underwent a Bentall procedure and total aortic arch replacement with an intraoperative stent inserted into the descending aorta. The deep hypothermic circulatory arrest time was 20 min. The clamping time was 169 min and the cardiopulmonary bypass time was 292 min. On the first postoperative day, the echocardiography showed the left ventricular ejection fraction was 60%. The patient was discharged safely 14 days later without any complications.

This report was approved by the research ethics committee of Guangdong Provincial People's Hospital and informed consent was obtained from the patient.

COMMENT

As a non-invasive examination, coronary computed tomography angiography is the preferred inspection method for evaluating CAD in patients with acute type A aortic dissection. However, coronary computed tomography angiography may be insufficient to quantify significant stenosis in some patients or unfeasible in cases of critical hemodynamic conditions (3, 4). Furthermore, conventional coronary angiography is often a high-risk procedure in patients with acute type A aortic dissection (8). Therefore, we introduced this technique for coronary visualization in patients with acute type A aortic dissection. This technique may prevent the extension of dissection and aortic rupture during preoperative coronary angiography and help determine whether if the patient requires a coronary artery bypass graft. What's more, this open heart technique can be done safely whilst the patient is on cardiopulmonary bypass with little time delayed which differs significantly from other approaches (MDCT and conventional coronary angiography). In our opinion, intraoperative open-heart coronary angiography is safe and effective in patients with acute type A aortic dissection.

DATA AVAILABILITY STATEMENT

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

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Research Ethics Committee of Guangdong Provincial People's Hospital. The patients/participants provided their written informed consent to participate in this study. Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

AUTHOR CONTRIBUTIONS

JY and XL wrote the main manuscript text. ZC and TS prepared figures. RF and CY were the managers of the whole study. All authors reviewed the manuscript.

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CASE REPORT
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Case Report: Simultaneous Ascending Aortic Dissection and Pulmonary Artery Dissection Combined by Aortopulmonary Fistula After Aortic Valve Replacement

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Aortopulmonary fistula with/without pulmonary artery dissection is an extremely rare and fatal complication of acute aortic dissection and is often discovered postmortem. We present a case with a simultaneous ascending aortic dissection and pulmonary artery dissection combined by aortopulmonary fistula after aortic valve surgery. However, the patient died of postoperative complications after surgery. Herein, the anatomical basis for this rare entity and its outcome is explored with an emphasis.

Keywords: aortic dissection, pulmonary artery dissection, aortopulmonary fistula, cardiac reoperation, surgical repair

INTRODUCTION

Acute aortic dissection (AAD) is rare with an incidence of 2.6–3.5 cases per 100,000 person-years (1) and so is pulmonary artery dissection (PAD) with only 150 cases reported from 1842 to 2018 (2). When a patient develops both two entities, it is extremely a rare and lethal condition, in particular, ascending aorta involvement [Stanford type A aortic dissection (TAAD)]. There is often some anatomical connection between the simultaneous appearance of TAAD and PAD including patent ductus arteriosus, aortopulmonary fistula, and aortopulmonary window (3–5). In addition to these characteristics, this case was secondary to aortic valve replacement, which was almost not reported in the previous studies.

CASE PRESENTATION

A 53-year-old man presented with acute chest pain and shortness of breath for 12 h was admitted to our hospital. He had a history of hypertension and diabetes mellitus and underwent mechanical aortic valve replacement for hypertension associated chronic severe aortic valve regurgitation 18 months ago. The blood tests on admission are shown in **Table 1** and most laboratory parameters deviated from the normal range. The bedside echocardiography confirmed the normal function of the mechanical aortic valve with an ejection fraction of 50%. The patient was examined by CT angiography (CTA) after he was stabilized by medical treatment and it showed a simultaneous ascending aortic dissection extending to descending aorta and PAD combined by aortopulmonary fistula (**Figure 1**).

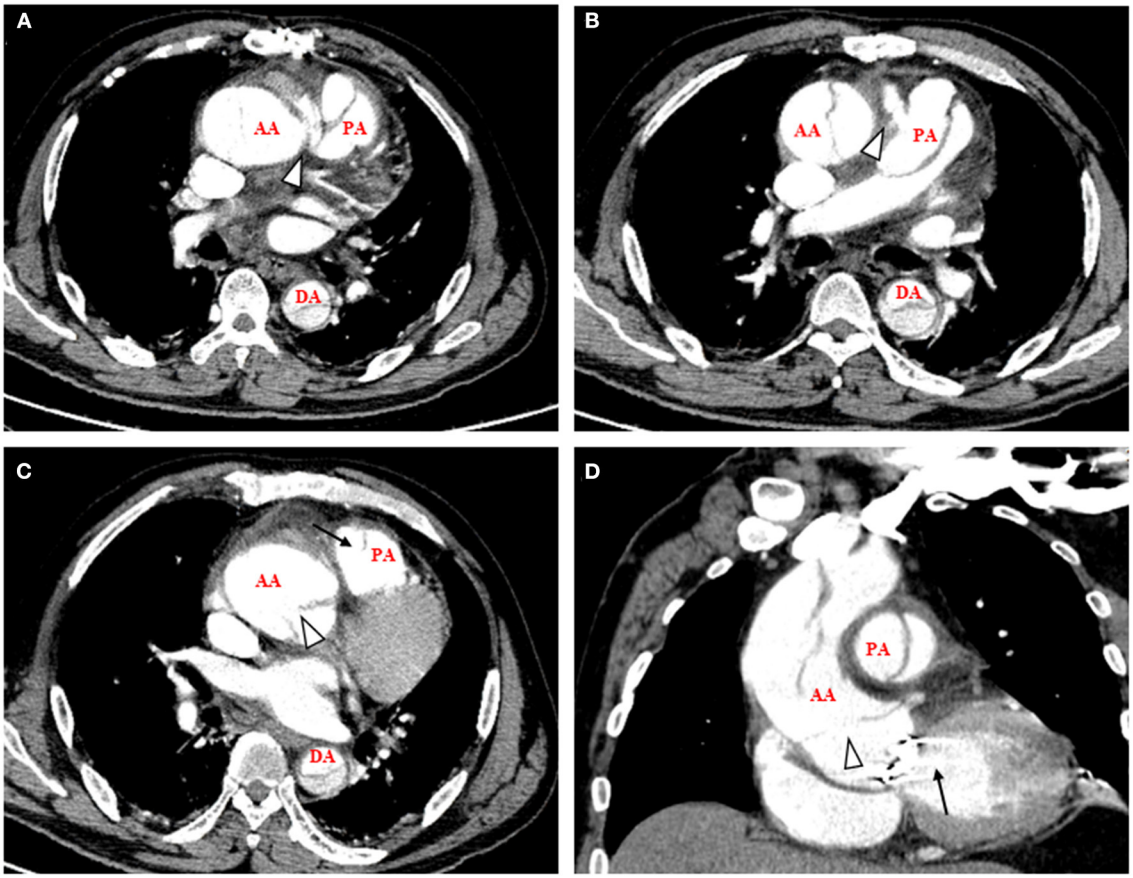

FIGURE 1 | CT angiography on admission showed a simultaneous ascending aortic dissection and pulmonary artery dissection combined by aortopulmonary fistula. **(A)** Axial view, arrowhead showed the aortopulmonary fistula near the aorta root. **(B)** Axial view, arrowhead showed the aortopulmonary fistula near the PA. **(C)** Axial view, arrowhead showed the intimal flap of AA and arrow showed the intimal flap of PA. **(D)** Sagittal view, arrowhead showed the intimal flap of AA that was in the aorta root and arrow showed the artificial aortic valve. AA, ascending aorta; PA, pulmonary artery; DA, descending aorta.

After we informed the families of the patient regarding the different treatments and related risks, including coronary artery involvement in aortic root dissection, renal malperfusion, heart failure, and especially complications associated with secondary cardiac surgery, the families and patient demanded surgical treatment.

Eventually, surgery was performed with median sternotomy and cardiopulmonary bypass was established *via* the intubation of the right atrial and right axillary artery, which was also used for selective cerebral perfusion in aorta arch replacement. The aorta root replacement and proximal anastomosis were performed first combined by removing the coronary arteries and reattaching them on the Dacron graft; in the meantime, the aortopulmonary fistula was closed with continuous 5-0 monofilament suture. Then, the hemiarch replacement and open distal anastomosis were completed under deep hypothermic circulatory arrest (nasopharyngeal temperature: 24°C). The surgery time was 474 min, cardiopulmonary bypass time was 185 min, cross-clamp time was 79 min, and deep hypothermic circulatory time was 23 min. The patient was transferred to the intensive care unit for further treatment and monitoring postoperatively. In the early period, there were no signs of unstable hemodynamics, respiratory insufficiency, and neurological disorders, and the ventilator weaning was performed 2 days after surgery. However, the patient was reintubated 10 days post-operatively due to dyspnea and methicillin-resistant Staphylococcus aureus was detected in sputum culture. Aspergillus was also detected in sputum culture 17 days postoperatively. Although vancomycin and fluconazole were used, the patient died of infectious shock 1 month after surgery.

DISCUSSION

According to the study by Jelani and Nosib (6), ascending aortic dissection could rarely lead to aortopulmonary fistula, which had been reported with 17 cases (13 acute and 4 chronic) until 2021 and further progression to PAD was an exceedingly rare condition. A case from Lempel et al. proposed a hypothesis for the etiology of the complex lesion (7). Ascending aorta and pulmonary artery trunk had a common adventitia, then

TABLE 1 | Primary blood tests of the patient on admission.

Variables	Value	Normal range
Leukocytes ($\times 10^9$/L)	10.59	3.50–9.50
Neutrophils ($\times 10^9$/L)	9.42	1.80–6.30
Hemoglobin (g/L)	129	130–175
Platelets ($\times 10^9$/L)	91	125–350
Prothrombin time (s)	19.7	11.5–14.5
D-dimer (μg/mL)	6.83	<0.50
Alanine aminotransferase (U/L)	45.0	4.0–41.0
Estimated glomerular filtration rate (ml/min/1.73 m^2)	32.7	>90.0
Creatinine (μmol/L)	188.0	59.0–104.0
Potassium (mmol /L)	5.23	3.50–5.10
Procalcitonin (ng/mL)	0.78	<0.50
Brain natriuretic peptide (pg/mL)	3,515	5–121
Cardiac troponin T (ng/mL)	1.88	<0.028

extravasation of blood from aorta would flow to the adventitial space of pulmonary artery and cause compression, and finally invaded the pulmonary artery (7). Although it is difficult to confirm this process currently, there is often a channel between the aorta and pulmonary artery in previous cases of TAAD and PAD (3–5), which suggests that the hemodynamic factor of the formation of PAD came from the aorta in the complex entities.

It was reported that the known aortic valve disease and recent aortic manipulation were risk factors of AAD (8), which indicated the damage and pathological remodeling in the aortic wall. Khatchatourian and Vala also reported that a patient with TAAD was found PAD in the routine postoperative CTA after emergency surgery (9). Therefore, the history of aortic valve replacement, in this case, might be related to this series of entities and reminded us of careful examination of the area around the aorta regardless of the level of image or surgery in the patient with TAAD with a history of aortic manipulation.

In the early years, most patients with TAAD combined with aortopulmonary fistula and/or PAD died and the outcomes improved in recent years (6). The total in-hospital mortality of the 13 acute cases was 38.5% (6). According to data from the International Registry of Acute Aortic Dissection (IRAD), surgical mortality of TAAD declined from 25 to 18% in the late 1990s to early 2010s (10). If PAD also existed, a preoperative acute left-to-right shunt and prolonged cardiopulmonary bypass time caused by the additional intraoperative process might affect the outcome. Besides, our case had a previous history of aortic valve replacement and cardiac reoperation undoubtedly posed a great challenge to the treatment. Nevertheless, surgery was the only definitive treatment for this entity currently and we undertook an emergency operation, while the function of the vital organs of the patients was deteriorating.

It was interesting that in a review with 150 cases with PAD, medical management alone achieved a success rate of almost 70%, while surgery alone achieved a success rate of almost 90%. Meantime in a case series, three patients developed PAD induced by pulmonary arterial hypertension and survived for nearly 10 years by medical management (11). Therefore, it suggests that the prognosis of the patients with TAAD and PAD mainly depends on the progress of management of TAAD.

In conclusion, TAAD with concomitant aortopulmonary fistula and PAD is an extremely rare and dangerous entity. This complex entity may be associated with the previous aortic manipulation. Emergency surgery is needed as the management of TAAD and the outcome of the patients also mainly depends on the treatment effect of TAAD currently.

DATA AVAILABILITY STATEMENT

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

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Ethics Committee of the Tongji Hospital of Tongji Medical College of Huazhong University of Science and Technology, written informed consent was obtained from the patient's legal guardian/next of kin for the publication of any potentially identifiable images or data included in this article.

AUTHOR CONTRIBUTIONS

LS drafted the manuscript and responsible for the collection of data and analysis. SX and JL designed the study and revised the manuscript. All the authors read and approved the final manuscript.

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CASE REPORT
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Case Report: Reconstruction of the Right Atrium With the Left Atrial Appendage

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We herein present a case of infective endocarditis of the mitral valve and a paravalvular abscess around the tricuspid valve. Preoperative blood culture confirmed the presence of pathogenic diphtheroids. During the operation, an unexpected infection of the free wall of the right atrium (RA) near the tricuspid annulus was found. We harvested the left atrial appendage (LAA) en bloc. After resection of the infected and abnormal tissues, the resected LAA was used to reconstruct the RA. The infected mitral valve was replaced with a mechanical valve without any accident. Postoperative echocardiography showed that the RA had a supple shape, with no kinking.

Keywords: infective endocarditis, right atrium, left atrial appendage, heart surgery, infection

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INTRODUCTION

Right-sided infective endocarditis rarely causes invasive complications such as abscesses or pseudoaneurysms (1). This can be explained by the lower pressure and lesser amount of muscle tissue around the tricuspid valve (TCV). However, the right atrium (RA) can become infected or can be invaded by tumors. In this case, the RA can be reconstructed using autologous pericardium, bovine pericardium, or an extracellular-matrix patch (ECM) (2). Herein, we present a case of paravalvular abscess around the TCV. We resected the infected RA and reconstructed it with the left atrial appendage (LAA).

CASE DESCRIPTION

A 25-year-old female patient presented to the Cardiac Surgery Department with intermittent fever and dyspnea that had lasted 6 months. She had no history of intravenous drug use. On the initial visit to Cardiac Surgery, her body mass index (BMI) was 25.1. Her respiratory rate was normal, but she had hypertension (blood pressure [BP] 180/90 mmHg). The electrocardiogram showed sinus rhythm. This patient had a moderate systolic murmur at the apex of the heart. She received telmisartan to control her BP. Serum creatinine (sCr) was 157 μmol/L. Her post-admission blood culture was positive, confirming that Corynebacterium diphtheriae was the pathogen. Transthoracic echocardiography (TTE) showed multiple small vegetations on the surface and tip of the mitral valve, mitral valve regurgitation, and mild TCV regurgitation (**Supplementary Video 1**). The primary diagnoses were infective endocarditis and renal dysfunction. TTE, computed tomography, and magnetic resonance imaging showed no involvement of the right atrium.

After broad-spectrum empirical antibiotic treatment (0.5 g imipenem/cilastatin 3 times a day, and 0.5 g vancomycin 2 times a day) for 1 week, the patient had no fever but was suffering from progressive heart failure. We performed transthoracic echocardiography again and found that the vegetation was stable, but the mitral regurgitation had progressed. To avoid progressive

heart failure, we decided to perform early mitral valve replacement using a median sternotomy. Cardiopulmonary bypass (CPB) was established in the standard manner. We cross-clamped the aorta, and we arrested the heart using antegrade cold del Nido cardioplegia. The appearance of the RA during the operation was normal (**Supplementary Video 2**). After the RA was opened, the free wall thereof near the tricuspid annulus was found to contain infected tissue and to have formed abscesses. Part of the tricuspid annulus was also infected. We first attempted to remove the partially infected tissue and then examined the LAA, which we harvested en bloc. The left atrium looked normal without any infection, so we did not conduct tissue culture or histological examination. We found three yellow and dark-red vegetations (about 1 × 1 cm each) on top of the mitral valve, which was also infected. We replaced this valve with a mechanical valve (size #25; St. Jude Medical [Abbott Laboratories, Abbott Park, IL, USA]). After resecting all infected and abnormal tissues of the RA free wall, we observed insufficient RA tissue to permit a safe closure, so we instead performed circumferential reconstruction of the RA. The area of infected tissue was about 4 × 3.5 cm, and it was close to the inferior vena cava and right atrioventricular groove (**Figure 1**; **Supplementary Video 2**). Part of the infected tricuspid annulus was also removed during the operation. We sutured the remnant tricuspid valve and annulus with a 5-0 PROLENE suture (Johnson & Johnson Medical Devices, New Brunswick, NJ, USA). The resected LAA was used to reconstruct the RA from the atrioventricular junction to the inferior vena cava using another 5-0 PROLENE suture. The autologous atrial tissue was pliable and easily handled. The saline test showed mild TCV regurgitation.

The patient was weaned off CPB successfully with no inotropic support. No bleeding occurred at the suture lines of the reconstructed RA. CPB time was 105 min, and aortic cross-clamp time was 75 min. TTE performed immediately after surgery showed that the reconstructed RA had a good shape (**Figure 2A**). Postoperative chest drainage was 235 mL over a 24-h period; drainage tubes were removed on day 2 post-surgery. The patient made a good recovery and was discharged 14 days post-surgery after standard antibiotic therapy.

Histology of the resected RA confirmed myocardial tissue infection and inflammation. Diphtheroids were cultured from the removed RA and mitral valve. At 6 months post-surgery, TTE showed good remodeling of the RA without kinking or shrinkage due to fibrosis (**Figure 2B**; **Supplementary Video 1**). Heart function was good. One month after surgery, the patient returned to work. Over 9 months of follow-up, she has had no fatigue or dyspnea.

DISCUSSION

We herein present a rare case of insufficient cardiac tissue for an attempt at directly closing the remnant RA. Selecting suitable tissue to reconstruct the RA is difficult. Previous studies have reported the use of different patches, such as

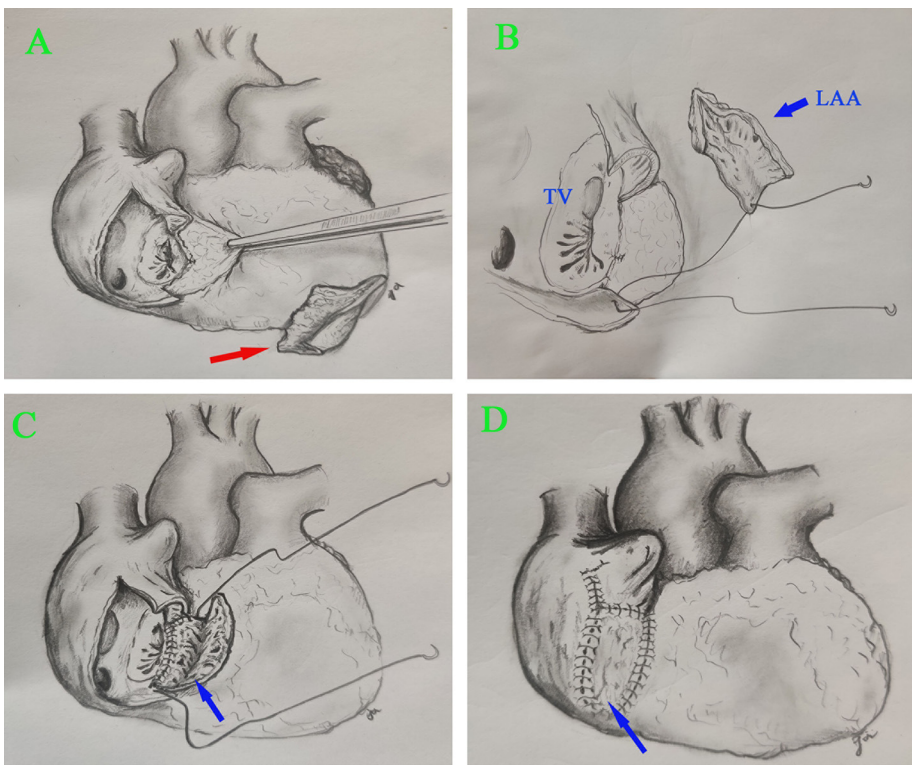

FIGURE 1 | Surgical view of the reconstructed RA. **(A)** Resection of infected RA tissue. **(B,C)** RA reconstruction using autologous LAA. **(D)** The reconstructed RA. Right arrow, infected RA tissue; blue arrow, LAA.

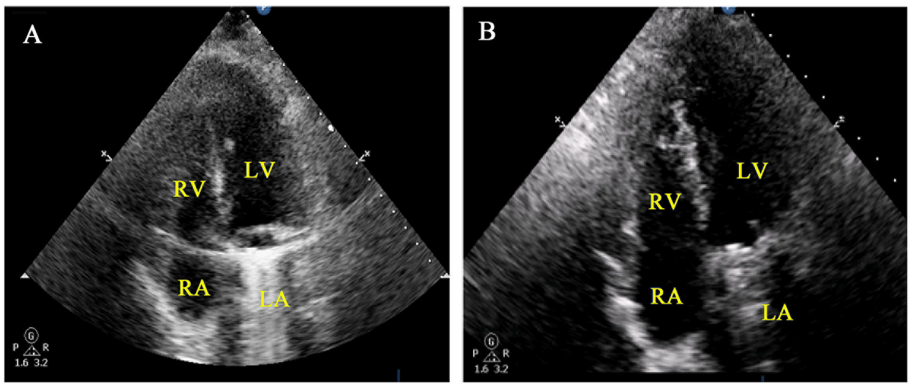

FIGURE 2 | Post-operative findings in a patient with infective endocarditis and paravascular abscess of the RA. **(A)** Immediately postoperative TTE of the RA and TCV. **(B)** TTE showed no shrinkage of the reconstructed RA 6 months after surgery.

porcine and bovine pericardia (2, 3), but such exotic tissues can cause an inflammatory reaction as well as fibrosis and scar formation. Fibrosis or scars in atrial tissue may be the source of arrhythmia because it promotes conduction slowdown, blockage, and reentry. ECM might be a better choice, but it is expensive and not available in developing countries (4). In addition, it is reputed to be biologically degradable. The autologous pericardium is a better choice for the construction of the RA after tissue excision.

We speculate that using autologous atrial tissue might have several advantages. Increasing the compatibility between the grafted and original tissues can prevent some scar tissue formation. The LAA is softer and more flexible than bovine or porcine pericardial tissue. Bleeding is more easily prevented when the patches have good adaptive margins with the surrounding remnant RA tissue. In this case, we needed to reconstruct the RA tissue around the right atrioventricular groove from the endocardium to the epicardium. The shape of the LAA on two planes was suitable for the reconstruction of the RA. The removed LAA is alive and maybe infection resistant and non-thrombogenic. An autologous graft does not induce an immunologic response. The use of autologous tissue also preserves the capacity to self-repair, adaptively remodel, and grow (5).

The LAA contributes up to 8–9% of left atrial volume and is highly variable in its morphology. It is also known to have an important effect on the release of atrial natriuretic peptide (ANP). The role of LAA in triggering and maintaining atrial fibrillation (AF) has received increasing attention, especially in patients with persistent AF or AF recurrence after repeated ablation (6). The complex anatomy and cellular structure of LAA may also predispose to arrhythmia because it promotes conduction slowdown, blockage, and reentry (7). In a retrospective study of 987 patients undergoing repeat catheter ablation for AF, 27% were found to have triggers in LAA (6). Not only is LAA a significant source of AF, but that electrical isolation or resection may have an important role in the treatment of persistent AF to avoid AF recurrence. Thrombosis is more likely to occur in the LAA than in other parts of the left atrium. Patients with left ventricular dysfunction or elevated left ventricular end-diastolic pressure might also be at risk of LAA thrombosis, even without AF (8). In our case, it was appropriate to remove the LAA and preserve its endocrine function in the reconstructed RA. However, the continuation of the endocrine function of the LAA after reconstruction still needs further exploration.

A positive blood culture confirms that Corynebacterium diphtheriae is the pathogen causing severe endocarditis in this case. Corynebacterium diphtheriae endocarditis is considered a rare disease that affects heart valves and seems to be very virulent and destructive. It is possible to select patients requiring emergency surgery based on the underlying valve pathology. Patients with abnormal or prosthetic valves should be prepared for emergency surgery if necessary (9). Surgery must include extensive debridement of all infected tissue, similar to that necessary for *staphylococcal endocarditis*.

Reconstruction of the RA carries the risk of several complications: bleeding, deformity, kinking, and degeneration. Our method is simple and economical, avoiding such complications. The size of the LAA is the main limitation of this method.

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 human participants were reviewed and approved by China Medical University. The patients/participants provided their written informed consent to participate in this study. Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

AUTHOR CONTRIBUTIONS

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

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fcvm.2021.782235/full#supplementary-material

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CASE REPORT
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Case Report: Management of a 10-Year-Old Patient Who Presented With Infective Endocarditis and Stanford Type A Aortic Dissection

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A 10-year-old girl presented with a chief complaint of cyclic vomiting since the last 12 h and chest pain since the last 6 h. She was diagnosed with Stanford type A aortic dissection. Intraoperatively, the aortic valve was found to be bi-lobed, and infective endocarditis associated with aortic valve perforation and rupture of the aortic sinus aneurysm, was also observed. Therefore, she underwent aortic valve replacement due to an enlarged aortic root and aortic sinus repair. The perioperative recovery was good. A large amount of bloody pericardial effusion was found in this child pre-operatively. Therefore, early surgical intervention was necessary. Acute aortic dissection rarely occurs in children. There are no clinical guidelines for the management of pediatric aortic dissection. However, if a large pericardial effusion exists, emergency surgery is necessary and effective. The treatment of the valve should be based on the actual situation. It is best to give priority to valve molding, although valve replacement is required in the majority of cases for infective endocarditis.

Keywords: pediatric acute aortic dissection, aortic valve replacement, infective endocarditis, aortic sinus aneurysm ruptured, Stanford type A aortic dissection

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INTRODUCTION

Aortic dissection (AD) mostly occurs in adults over 50 years of age, with a high fatality rate (1); it is rare in children. Based on the statistical data of 12,142 patients with AD in New York State from 1996 to 2005, no children under the age of 15 years were found, and only 45 patients under the age of 21 years were found (2). Hua et al. (3) and Shamszad et al. (4) reported that the age of onset in children was mostly 0∼5 years and 15∼20 years, most of them were male, and case fatality rate was 3∼17%. There is no relevant report in China. Abroad, the youngest patient reported was a 1-day-old (5). Here, we report a case of successful surgical management of infective endocarditis with ruptured aortic sinus aneurysm resulting in acute AD.

This study was approved by the Institutional Review Board of Shengjing Hospital of China Medical University. The patient's parents provided written informed consent for publication of the data and associated images.

CASE DESCRIPTION

A 10-year-old girl presented to our hospital with headache and nausea of 12 h duration. Physical examination revealed the following: heart rate of 120 bpm, blood pressure of 104/59 mmHg, respiratory rate of 24 breaths/min, and temperature of 36.2°C. Auxiliary examination including complete blood count revealed the following: leukocyte count, 28.85 × 10^9/L; neutrophil count, 88.4%; lymphocyte count, 7.6%; hemoglobin level, 107 g/L; and platelet count, 401 × 10^9/L. Troponin I level (high sensitivity) was 0.0619 ug /L, myoglobin level was 12.7 ug/L, and isoenzyme mass was 0.9 ug/L. Blood biochemistry revealed the following: total protein level, 62.1 g/L; albumin level, 34.5 g/L; alanine aminotransferase level, 26 U/L; aspartate aminotransferase level, 63 U/L; blood-glucose level, 13.83 mmol/L; creatine kinase level, 103 U/L; and creatine kinase MB isoenzyme level, 20 U/L. C-reactive protein level was 11.50 mg/L; prothrombin time, 13.8 s; prothrombin time activity, 71%; prothrombin normalized ratio, 1.3; prothrombin ratio, 1.3; activated partial thrombin time, 30 seconds; fibrinogen content, 3.1 g/L; thrombin coagulation time, 16.7 seconds; and D-dimer level, 481 ug/L. Three hours later, the patient vomited again, and began experiencing tenderness in the region of the chest anterior to the heart. Electrocardiography showed sinus tachycardia and S-T segment elevation. The patient vomited frequently, and was pale and tachypneic, with low-pitch heart sounds; her pulse pressure was increased. She was immediately transferred to the pediatric intensive care unit for further treatment, where she was sedated and given antiemetics. Physical examination revealed audible diastolic murmurs in the auscultation area of the aortic valve. On day 2, Color Doppler echocardiography revealed Stanford type A: Debakey I AD, aortic regurgitation, and massive pericardial effusion; the aortic root was widened to 50 mm in diameter (**Figure 1**). Computed tomography (CT) revealed a widened mediastinum, significant dilation of the posterior part of the aortic root (∼58 mm at its widest point) extending up to the aortic arch. The patient had neither the exit tear, nor the involvement of the supra-aortic branches and the involvement of the abdominal aorta branches. Two lacerations could be seen above the aortic valve ring. There was blood flow through the breach from the true to the false lumen (**Figures 2**, **3**). Consequently, on day 3, we decided to perform ascending aortic replacement. The patient was placed in the supine position, and a midsternal incision was made. Total intravenous anesthesia was administered, and cardiopulmonary bypass (in which a venous catheter was inserted into the superior and inferior vena cavae and an arterial cannula was placed in the ascending aorta) was performed. When the pericardium was opened, a large amount of hemorrhagic effusion was visible, and a huge aneurysm with a diameter of 5 × 4 cm could be seen in the posterior wall of the aortic root (**Figure 4**). Due to great tension, the boundaries of the aneurysm and posterior wall of the aortic root were unclear, and tissue adhesion continued upward to the junction between the ascending aorta and right pulmonary artery, causing left atrium compression. After incising the ascending aorta, bicuspid aortic valve (BAV) malformation was observed with fusion of the right and non-coronary cusp and perforation of the left coronary cusp, along with many vegetations attached to the surface. Two breaches, with diameters of 2 and 3 mm, were observed at the midpoint of the non-coronary sinus, which communicated with the aortic root aneurysm, and the surfaces of the breaches were rough with a few neoplasms visible (**Figure 5**). The damaged aortic wall, infected aortic valve, and neoplasms were removed with scissors, and iodophor cotton balls were used for disinfection. The aortic root was small and needed to be widened to allow for implantation of an adult-size valve. We decided to use the Konno method to enlarge the aortic valve ring. The aortic wall and valve ring were cut down along the right coronary sinus and left inferior valve lobe triangle until the ventricular septum was 1 cm from the aortic valve ring below the posterior wall of the right ventricular outflow tract and the free wall of the right ventricle. The artificial vascular patch was trimmed to an appropriate size and shaped like a tear drop. The narrowest part of the tear drop was placed on the left ventricular surface. The 4-0 line was passed through the patch, which was continuously sutured upward on the left ventricular surface and ascending aortic wall to enlarge the valve ring and ventricular septal incision. We repaired the tear in the aorta with the artificial vascular patch (**Figure 6**). After widening the suture, the size of the valve ring was measured and the 18# St. Jude mechanical aortic valve was used for the replacement. The valve was sutured with valve replacement suture at the normal position of the aortic valve ring, and the artificial valve was sutured with 4-0 spacer line at the position of the widened artificial vascular patch. After valve replacement, the left and right coronary openings were found to be unblocked, and the incision on the ascending aorta was closed with a 4-0-line double-layer mattress suture + continuous suture.

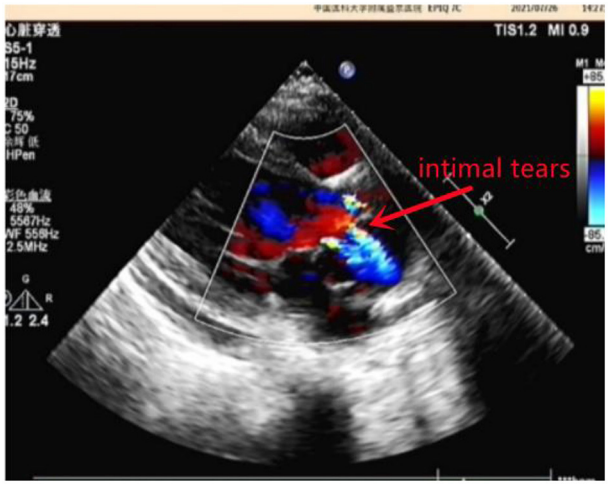

FIGURE 1 | Color Doppler echocardiography.

The following day, the patient was weaned off the ventilator. She left the ICU on post-operative day 3 and was discharged from the hospital on post-operative day 10. Follow-up CT 4 weeks after surgery showed that the false lumen had disappeared (**Figure 7**). The patient had no symptoms such as chest pain, respiratory distress, or fever; her post-operative international

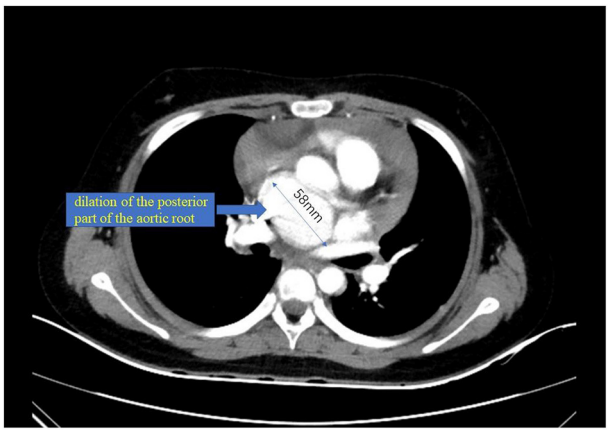

FIGURE 2 | Computed tomography (CT).

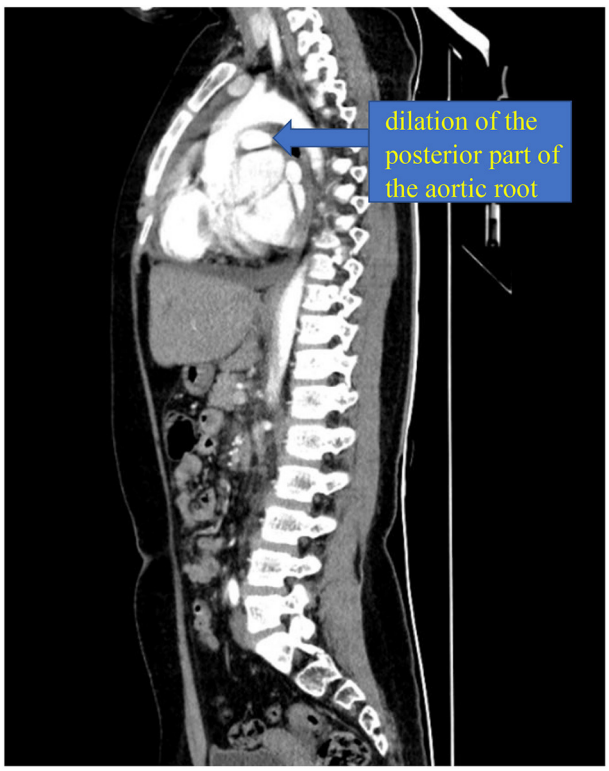

FIGURE 3 | Computed tomography (CT).

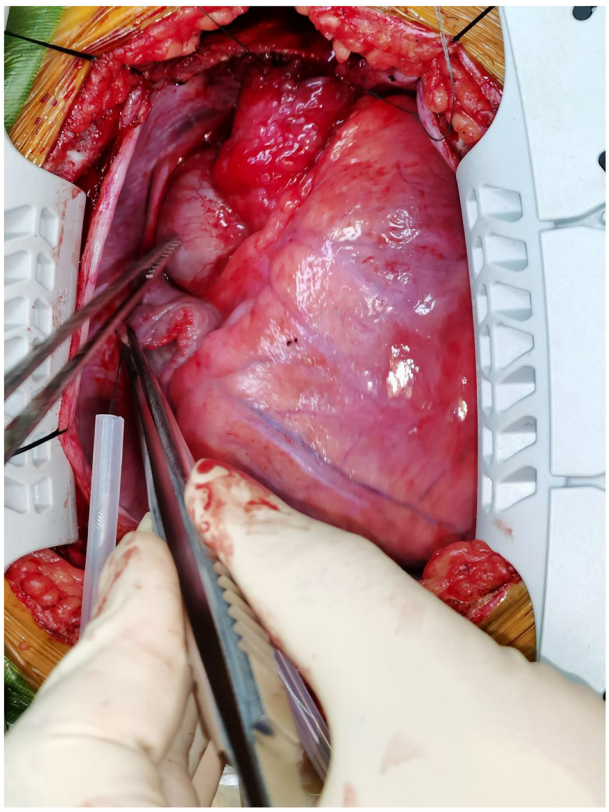

FIGURE 4 | The huge aneurysm.

normalized ratio was maintained at about 2.0, with no obvious bleeding tendency.

DISCUSSION

Acute AD is rare in pediatrics and can be easily misdiagnosed. A complete history and systematic physical examination are the most important parts of the assessment of children with chest pain. When the history and physical examination indicate that the pain may have a serious cause, the relevant auxiliary examinations should be urgently requested to identify the cause and determine the course of treatment. In this patient, precordium tenderness appeared 6 h after a sudden-onset headache and nausea; physical examination revealed an increase in heart rate and significant cardiomegaly. Based on the characteristics of the pain and abnormal examination findings, we considered the chest pain to be due to an organic disease and the possibility of a fatal cardiovascular disease to be high. A diagnosis of AD was made through our hospital emergency route. After the surgery, the patient's family was asked about her medical history, and they revealed that the child was hospitalized for encephalitis and septic shock 2 months ago, but no heart murmur was found at the time. After discharge, the child still had recurrent low-grade fever. It was believed that the insufficient course of anti-inflammatory treatment led to the spread of infection and colonization of the heart valve and aortic sinus by bacteria, leading to the occurrence of AD. The patient did not have hemoculture at the time. Hemocultures before and after the surgery also did not find any microbe responsible.

AD is a tear between the intima and media of the aorta due to various reasons. The intima and media of the aorta are separated and blood flows in, resulting in the formation of a true and false lumen. A typical AD shows a septum or an inner diaphragm between the true and false lumens. True and false lumens can be

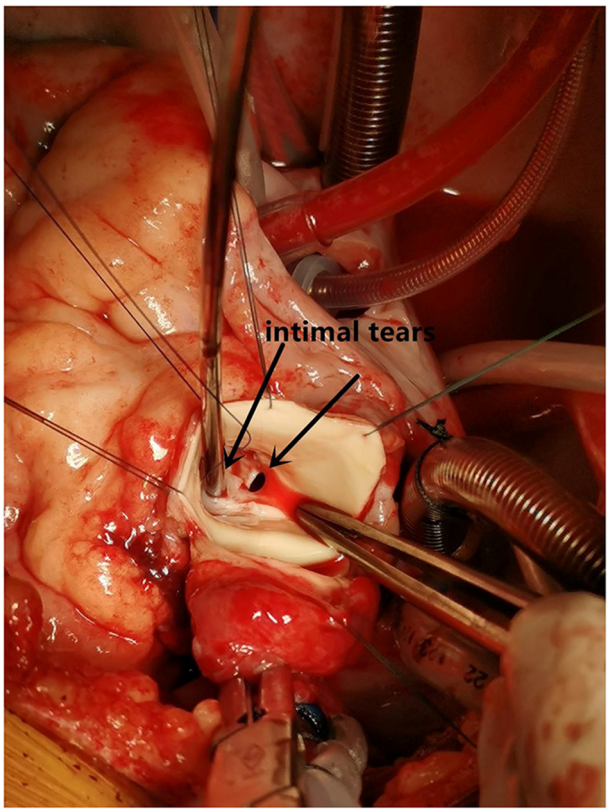

FIGURE 5 | The intimal tears.

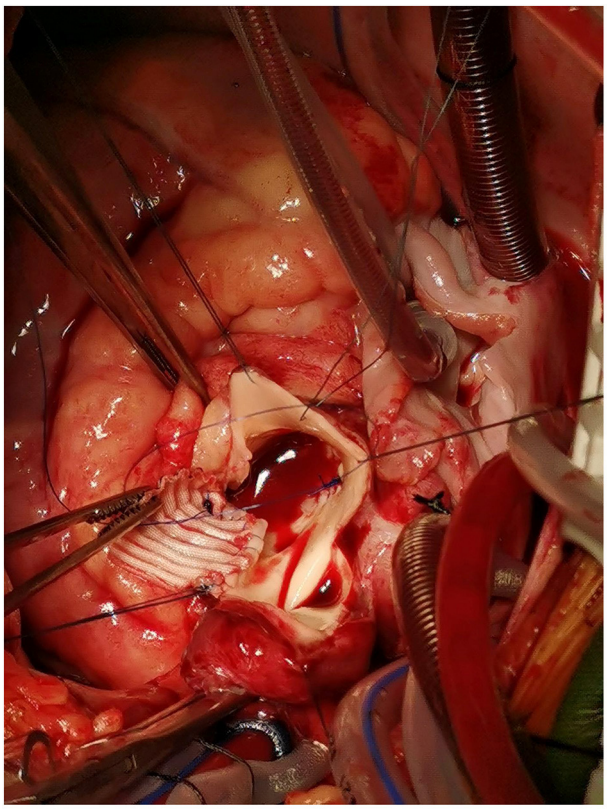

FIGURE 6 | The artificial vascular patch.

communicating or non-communicating. Blood can flow between the true and false lumens or form a clot (1). Hypertension and atherosclerosis are associated with 90% of adult cases of AD, while congenital and genetic diseases, such as congenital aortic stenosis, Ehlers-Danlos syndrome TYPE IV, and Marfan syndrome among others, are common in children or adolescents with AD. It can also be seen in patients with trauma, infection, and drug use.

Chest pain is a common complaint in children. Although it is mostly non-organic, a serious and life-threatening etiology may be present. The challenge for physicians is identifying the few patients with severe etiologies as early as possible. A complete history and systematic physical examination are the most important parts of the assessment in children with chest pain. When the history and physical examination indicate a potentially serious cause, auxiliary investigations should be urgently carried out to identify the etiology and initiate treatment. To the best of our knowledge, there are no clinical guidelines for the management of pediatric AD. Among conservative treatment, endovascular grafting, and valve replacement, the appropriate treatment strategy remains controversial. However, in pediatric patients, artificial blood vessels may not be the first choice due to the commercial unavailability of an appropriate graft and the problem of future growth of the artery. Therefore, in this case, we did not perform ascending aorta replacement; instead, we repaired the part of the aortic wall which was involved in the tear in the aortic intima and media. Post-operatively, the false lumen disappeared. It should be noted that this patient was suffering from BAV malformation; foreign studies have been found to be the second most common cause of heart disease in

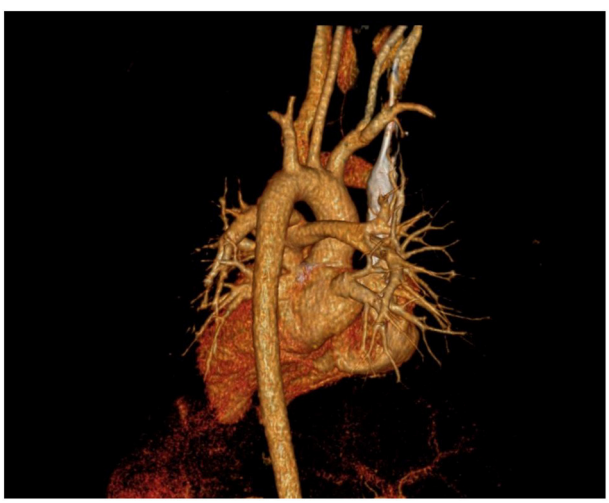

FIGURE 7 | Follow-up CT.

all children requiring valve replacement (6). BAV is also a major independent risk factor for AD in the Chinese population (1). When children with aortic valve disease need surgical treatment, valve repair is the first choice. If the valve cannot be repaired, or repair fails, an aortic valve replacement is compulsory. If infective endocarditis develops, mechanical valves are the best option, when available. Lifelong anticoagulant therapy is required after mechanical flap replacement. In children, the theoretical rate of anticoagulation-related complications should be higher due to lack of compliance and activity restrictions; however, the actual risk is lower in children than in adults. It has been reported in the literature that children have a 90–100% chance of avoiding thromboembolism and a 96–100% chance of avoiding bleeding during the 10 to 20-year follow-up period (7, 8). Due to age or disease, the aortic valve ring and root in children are often too small to allow for implantation of the minimum adult size (19 mm) valve. If the aortic valve ring is enlarged and the root widened at the same time, a larger prosthetic valve can be implanted and is conducive for reducing the transvalvular pressure gradient difference and improving heart function; it may also prevent the need to replace the artificial valve with a larger valve after growth and development of the child (9). In the Konno method, also known as aortic ventriculoplasty, the valve ring is fully exposed and can be expanded more effectively, while dredging the left ventricular outflow tract. However, it requires two patches, and the operation is complicated and difficult. In pediatric patients with narrower aortic annulus and root, and in some with left ventricular outflow tract stenosis, we prefer to use the Konno technique to enlarge the annulus, which allows for the implantation of larger valves. Post-operative follow-up showed that the child had a satisfactory recent effect. However, at the 3-month follow-up, the transprosthetic gradient reached 30 mmHg. Although the patient is currently asymptomatic and has good cardiac function, it is not certain whether she will require more surgery and how the cardiac function will be in the future; further observation and follow up are required

Since pediatric AD is uncommon and there are no clinical guidelines for its management, we should keep in mind that an early surgical approach with aortic replacement is sometimes necessary instead of conservative treatment or endovascular grafting.

DATA AVAILABILITY STATEMENT

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

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Ethics Committee of Shengjing Hospital of China Medical University. Written informed consent was obtained from the patient's family for the publication of any potentially identifiable images or data included in this article. Patient consent form was read and signed by the patient's parents.

AUTHOR CONTRIBUTIONS

YL participated in data collection, data analysis, and manuscript writing. MW participated in data collection and data analysis. NY participated in data analysis. LW participated in project development. DL participated in project development, data analysis, and manuscript writing. All authors read and approved the manuscript.

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frontiers in Cardiovascular Medicine

CASE REPORT
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Case Report: Three-Dimensional Printing–Assisted Surgical Treatment of Complex Body Vein Ectopic Drainage

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Introduction: Complex ectopic drainage of body veins is a rare congenital disease. Its preoperative diagnosis and surgical choice can be considerable challenges.

Case Summary: A 5-year-old patient was diagnosed precisely by preoperative transthoracic echocardiography, computed tomography (CT), three-dimensional (3D) reconstruction, and three-dimensional (3D) printing of the heart and great blood vessels. The operation was performed successfully using flexible intraoperative intubation strategies.

Conclusion: 3D printing technology can assist in the formulation of surgical protocols for complex body vein ectopic drainage. Flexible intubation strategies can increase the success of the operation.

Keywords: complex body vein ectopic drainage, 3D reconstruction, 3D printing, inferior vena cava, hepatic vein, intubation complex body vein ectopic drainage, intubation

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INTRODUCTION

Abnormal drainage of the inferior vena cava (IVC) into the left atrium (LA) is a rare congenital condition. Especially when associated with ectopic hepatic vein drainage and atrial septal defects, its preoperative diagnosis and surgical choice can be considerable challenges with no uniform standards (1, 2).

Here, we report a case of a patient with ectopic drainage of the inferior vena cava into the left atrium, ectopic drainage of the hepatic vein, and atrial septal defects. We precisely developed surgical protocol using 3D printing technology and successfully performed the operation of complex body vein ectopic drainage.

CASE REPORT

A 5-year-old boy with a history of allergy to milk and eggs experienced frequent colds. On physical examination, the patient was well-developed and showed no signs of hypoxia, such as cyanosis. He was found to have a grade 3/6 systolic ejection murmur between the second and third intercostal space along the left sternal border. Moreover, his blood oxygen saturation of the extremities was in the normal range. The patient underwent a detailed examination before

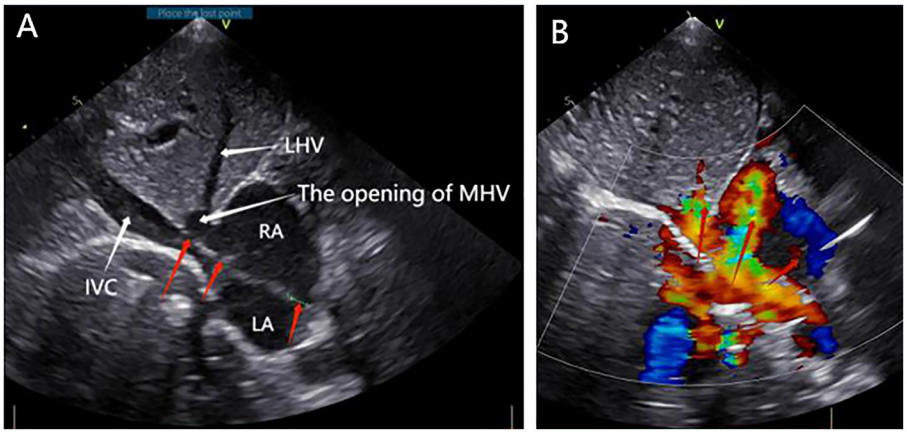

FIGURE 1 | Preoperative transthoracic echocardiography. **(A)** Right atrium (RA), left atrium (LA), left hepatic vein (LHV), the opening of middle hepatic vein (MHV), inferior vena cava (IVC) (white arrowheads), and three atrial septal defects (ASD) (red arrowheads). **(B)** Blood flowed from the left atrium to the right atrium (red arrowheads).

the operation including chest radiography, electrocardiography, transthoracic echocardiography, computed tomography (CT), three-dimensional (3D) reconstruction and 3D printing of the heart and great blood vessels, and detailed laboratory examination. The patient's chest radiograph showed a slight swelling of the pulmonary artery. Transthoracic echocardiography showed three secondary atrial septal defects, and that a part of the right hepatic vein and inferior vena cava drained into the left atrium (**Figures 1A,B**). The 3D reconstruction and 3D printing models showed the following abnormalities: there were three atrial septal defects (one of fossa ovalis type, one near the superior vena cava, and one near the inferior vena cava); the left hepatic vein joined the middle hepatic vein, and the opening was located in the right atrium (**Figure 2A**); the right hepatic vein drained into the inferior vena cava, and the opening of the inferior vena cava was located in the left atrium (**Figures 2B,C**). **Figure 2D** shows that two atrial septal defects could be revealed from the right atrium incision level.

During the establishment of cardiopulmonary bypass, routine aortic cannulation, superior vena cava cannulation, cold perfusion cannulation, and superior vena cava and aortic blockade were performed. Owing to the abnormality of the inferior vena cava opening, inferior vena cava cannulation was not performed. One right atrium drain and two left atrium drains were placed in the opening of the hepatic vein in the right atrium and the opening of the inferior vena cava in the left atrium through the atrial septal defect, respectively, for intracardiac blood drainage. The field was well-exposed, and the patient's blood perfusion and vital signs were good. The foramen ovale atrial septal defect was viewed directly, and the diameter was about 15 mm (**Figure 3**). The left hepatic vein joined the middle hepatic vein, and the opening was located in the right atrium (**Figure 3A**). During the operation, the interatrial septum was cut off, and we found the right hepatic vein draining into the inferior vena cava and the opening of the inferior vena cava in the left atrium next to the right inferior pulmonary vein (**Figure 3B**). A transverse septum, where the inferior vena cava met the left atrium, was found (**Figure 3B**). The atrial septal defect was closed by continuous suture of the bovine pericardium slice of the corresponding area with 5-0 prolene suture, and the openings of the hepatic vein and inferior vena cava were separated into the right atrium. The surgery was successful, and the patient was discharged from hospital on the fifth postoperative day. A postoperative echocardiogram performed on the third postoperative day demonstrated that there was no residual atrial shunt, and the blood supply to the heart from the IVC and the hepatic veins was corrected. At the 1-month follow-up, the patient was doing well.

DISCUSSION

To our knowledge, there have been several cases of patients with inferior vena cava complicated with pulmonary venous drainage and atrial septal defect (1–3). This is the first case describing the detailed clinical features, preoperative examination data, and surgical data of a patient with inferior vena cava complicated with abnormal hepatic venous drainage and atrial septal defect. Our experience with our first case cautions surgeons who may encounter patients with ectopic drainage of inferior vena cava and hepatic vein.

Ectopic drainage of the inferior vena cava into the left atrium is usually associated with cyanosis (1–4). However, this patient did not have obvious signs of cyanosis and had good blood oxygen saturation of the extremities, which may be due to the large diameter of the atrial septal defect and the role of the transverse septum at the orifice of the inferior vena cava in the left atrium. Because the pressure in the LA is higher than that in the RA in the early stage of the disease, the septum carries most of the blood from the IVC into the RA through the ASD, and oxygenated blood in the pulmonary circulation is transported to the whole

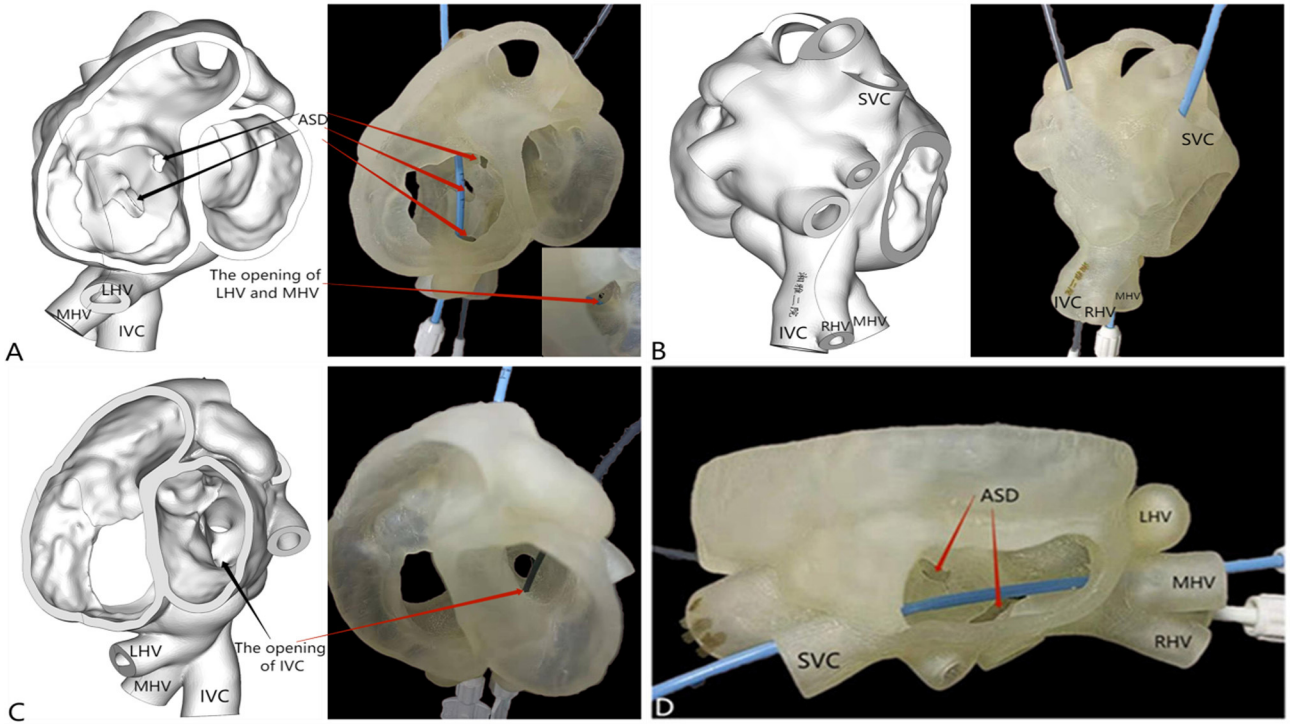

FIGURE 2 | 3D reconstruction and 3D printing. **(A)** Three large atrial septal defects (one of fossa ovalis type [diameter of 6 mm], one near the superior vena cava [diameter of 15 mm], and one near the inferior vena cava [diameter of 8 mm]); the left hepatic vein joined the middle hepatic vein, and the opening was located in the right atrium. **(B,C)** The right hepatic vein drained into the inferior vena cava, and the opening of the inferior vena cava was located in the left atrium. **(D)** Two atrial septal defects were visible from the right atrium incision level.

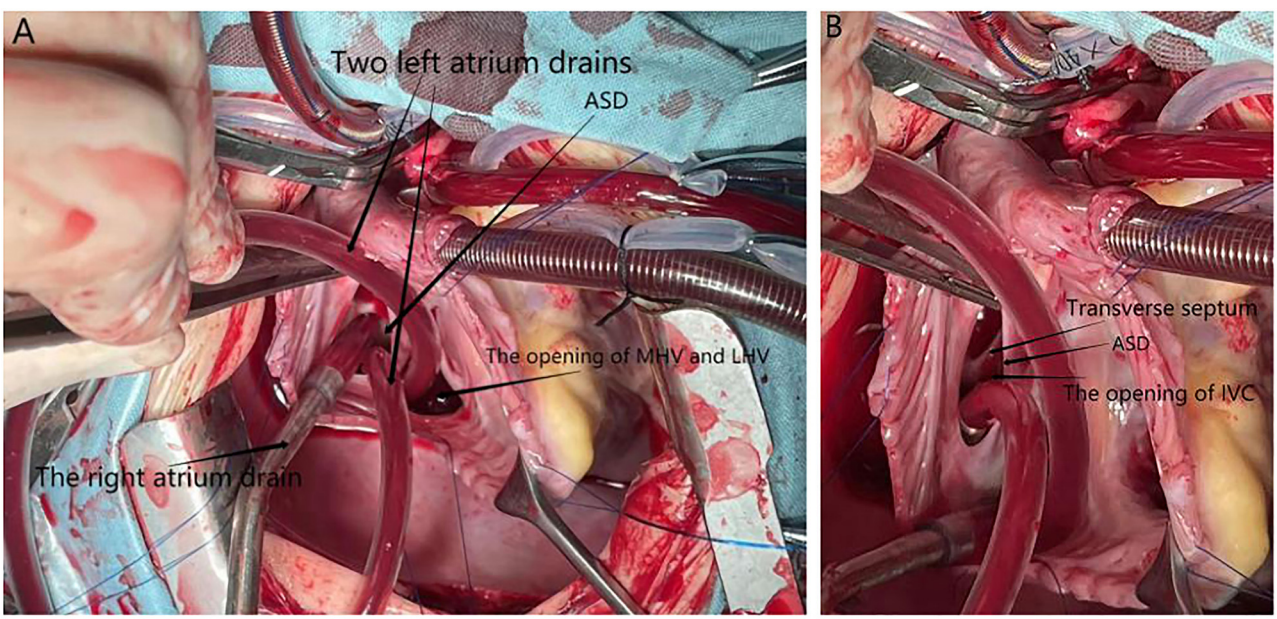

FIGURE 3 | An intraoperative photograph (after the RA was cut off). **(A)** ASD, the opening of LHV and MHV, two left atrium drains, and one right atrium drain are indicated by black arrowheads. **(B)** Transverse septum at the orifice of the inferior vena in the left atrium and the opening of the inferior vena cava are indicated with black arrowheads.

body. As the disease progresses, significant left-to-right shunting tends to lead to pulmonary hypertension. Increased pulmonary artery pressure would reduce left-to-right shunt; therefore, a large amount of blood from IVC would flow to the systemic circulation, thereby increasing symptoms of hypoxia such as decreased oxygen saturation. Thus, the patient needed treatment to prevent further progression of the disease.

On the basis of preoperative transthoracic echocardiography and CT examination of cardiovascular blood vessels accompanied with 3D reconstruction and 3D printing, we were able to visualize the patient's cardiac abnormalities well, laying a good foundation for the choice of surgical methods. CT imaging has many advantages in diagnosing and treating cardiovascular diseases, with clearer images and faster scanning speed. 3D printing models can reflect complex cardiovascular malformations more intuitively and accurately (5). We clearly observed the abnormal drainage of the inferior vena cava and hepatic veins through 3D reconstruction and 3D printing model. 3D imaging aids in surgical planning and reduces redundant surgical interventions. In addition, it may also help to reduce the rate of misdiagnosis and surgical errors (6).

Since the inferior vena cava cannulation was not performed during the establishment of cardiopulmonary bypass, the three atrium drains placed in the left atrium and the right atrium fully exposed the surgical field of view and facilitated the operation, indicating that cannulation and occlusion of the inferior vena cava do not need to be performed when there is a way to drain the blood from the inferior vena cava during the establishment of cardiopulmonary bypass to guide operations in the future.

CONCLUSION

This is the first case of the ectopic drainage of the inferior vena cava into the left atrium with ectopic drainage of the hepatic vein and atrial septal defects. 3D printing technology can assist in the formulation of surgical protocols for complex body vein ectopic drainage. Flexible intubation strategies can increase the success of 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/s.

ETHICS STATEMENT

Ethical review and approval was not required for the study on human participants in accordance with the local legislation and institutional requirements. Written informed consent to participate in this study was provided by the participant's legal guardian/next of kin. Written informed consent was obtained from the minor's legal guardian/next of kin, for the publication of any potentially identifiable images or data included in this article.

AUTHOR CONTRIBUTIONS

TZ, QW, GS, and SH were responsible for the diagnosis and treatment of the patient. LW collected clinical data and prepared the manuscript. All the authors have read and approved the final manuscript, have agreed to be accountable for the content of the work, contributed to the article, and approved the submitted version.

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ORIGINAL RESEARCH
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Midterm Outcome After Septal Myectomy and Medical Therapy in Mildly Symptomatic Patients With Hypertrophic Obstructive Cardiomyopathy

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Objective: The purpose of this study was mainly to determine the midterm outcome of septal myectomy (SM) and medical therapy (MT) in mildly symptomatic patients (NYHA class II) with hypertrophic obstructive cardiomyopathy (HOCM).

Methods: The study cohort consisted of 184 mildly symptomatic patients with HOCM evaluated in Beijing Anzhen Hospital, Capital Medical University between March 2001 and December 2017, including 82 patients in the SM group and 102 patients in the MT group. Overall survival and HCM-related survival were mainly observed.

Results: The average follow-up time was 5.0 years. Compared to patients accepting MT, patients treated with SM were associated with comparable overall survival (96.5% and 93.1% vs. 92.9% and 83.0% at 5 and 10 years, respectively; $P = 0.197$) and HCM-related survival (98.7% and 98.7% vs. 94.2% and 86.1% at 5 and 10 years, respectively; $P = 0.063$). However, compared to MT, SM was superior at improvement of NYHA class (1.3 ± 0.6 vs. 2.1 ± 0.5, $P < 0.001$) and mean reduction of resting left ventricular outflow (LVOT) gradient ($78.5 \pm 18.6\%$ vs. $28.3 \pm 18.4\%$, $P < 0.001$). Multivariate analysis suggested that resting LVOT gradient in the last clinical examination was an independent predictor of all-cause mortality (HR = 1.017, 95%CI: 1.000–1.034, $P = 0.045$) and HCM-related mortality (HR = 1.024, 95%CI: 1.005–1.043, $P = 0.012$) in the entire cohort.

Conclusion: Compared with MT, SM had comparable overall survival and HCM-related survival in mildly symptomatic HOCM patients, but SM had advantages on improving clinical symptoms and reducing resting LVOT gradient. Resting LVOT gradient in the last clinical examination was an independent predictor of all-cause mortality and HCM-related mortality.

Keywords: hypertrophic obstructive cardiomyopathy (HOCM), mild symptom, septal myectomy, medical therapy (MT), outcome

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INTRODUCTION

Hypertrophic obstructive cardiomyopathy (HOCM) is a genetic heart disease characterized by marked cardiomyocyte hypertrophy and left ventricular outflow tract (LVOT) obstruction (1–3). LVOT obstruction not only leads to exertional dyspnea, fatigue, chest pain, and limited exercise capacity (4), but also increases all-cause mortality and the incidence of sudden cardiac death (SCD) in patients with hypertrophic cardiomyopathy (HCM) (5, 6). Septal myectomy (SM) has been proven by multiple studies that provides excellent long-term survival and freedom from recurrent symptoms in highly symptomatic patients with HOCM (7, 8). Currently, SM is mainly recommended for HOCM patients with severe symptoms (NYHA class III-IV or recurrent exertional syncope) despite optimal medical therapy (MT) in European and American clinical practice guidelines (Class I indication) (9, 10). However, one study indicated that in mildly symptomatic (NYHA class II) or asymptomatic patients with HOCM, severity of LVOT gradient at rest was independently associated with a higher risk of developing heart failure and death (11). In our recent research, we have demonstrated that although alcohol septal ablation (ASA) did not provide better long-term survival in mildly symptomatic HOCM patients compared with MT, ASA could evidently improve clinical symptoms and reduce LVOT gradient, which may be a kind of reasonable alternative for patients intolerant to MT (12). Nevertheless, there is no definite evidence for SM applying to mildly symptomatic HCM patients with severe obstruction. Recently, a study made by Alashi et al. (13) suggested that in patients with HOCM, earlier surgery vs. surgery for guideline-based Class I indication had a higher long-term survival, which was similar to the age- and sex-matched US population. However, there is no study that directly compares the outcome of SM and MT in mildly symptomatic patients with HOCM. Therefore, this study was conducted to primarily evaluate the outcome of SM in mildly symptomatic patients with HOCM, as a comparison with MT.

MATERIALS AND METHODS

Study Patients

This retrospective study consisted of 184 mildly symptomatic patients with HOCM from Beijing Anzhen Hospital, Capital Medical University between March 2001 and December 2017, including 102 patients in the MT group and 82 patients in the SM group. Informed consent was obtained from each patient before the study began. This study was conducted according to the ethical standards of Helsinki Declaration, Chinese clinical practice regulations and guidelines, and rules of Medicine Ethics Committee of Beijing Anzhen Hospital (institutional review board number: No. 2020087x, date of approval: December 22, 2020). This study has been registered on the Chinese Clinical Trial Registry (No. ChiCTR2000041464). Each patient was adult (age $\geq$ 18 years) and had an established diagnosis of HOCM. In an adult, the diagnosis of HOCM was made as described formerly as follows (9, 10, 14): (1) the wall thickness of one or more left ventricular myocardial segments measured by any imaging technique was $\geq$15 mm and without any other disease accounting for cardiomyocyte hypertrophy; (2) LVOT gradient $\geq$ 30 mmHg at rest or after provocation caused by anterior systolic displacement of mitral valve. The MT group consisted of mildly symptomatic HOCM patients who have obtained optimal MT (maximum tolerable dose of beta-receptor antagonists and/or calcium channel blockers). Consecutive patients met the following criteria were enrolled in the SM group, including: (1) intolerant to medical treatments (After taking β-receptor antagonist or calcium channel blocker, the patient had obvious symptoms such as hypotension-related dizziness or evidently fatigue, which can significantly reduce the patients' quality of life); (2) had a strong wish for symptomatic relief; (3) LVOT gradient $\geq$ 50 mmHg at rest or after provocation. Patients met the following criteria were excluded from the SM groups: (1) patients with severe comorbidities, such as severe hepatic and/or renal dysfunction, malignant tumors; (2) patients with complete right bundle branch block; (3) patients with high risk of SCD, for example: recorded exertional syncope, family history of premature SCD and non-sustained ventricular tachycardia; (4) patients have been treated with ASA. All patients were informed about potential risk of SM and agreed with the procedure. Procedure of SM have been described in detail on several previous reports (15–19).

Follow-Up

In the MT group, follow-up started at the first clinic contact of patients after March 1, 2001 in Beijing Anzhen Hospital; in the SM group, follow-up started on the day of surgical intervention. If no endpoints occurred during follow-up, follow-up ended at the last check-up and the final censoring date was set at April 1, 2021. Follow-up was conducted by means of clinic visit, telephone contact and online communication. The following parameters were documented: symptoms, arrhythmic events and pacemaker implantation, causes of death (confirmed by reviewing the medical records and national registries of deaths or communicating with family members of the patients), electrocardiography and echocardiographic parameters.

Endpoints

The primary and secondary endpoints of this study were all-cause mortality and HCM-related death, respectively. In addition, we want to determined: (1) predictors of all-cause mortality and HCM-related mortality; (2) difference of symptom improvement, occurrence of new-onset atrial fibrillation (AF) and echocardiographic parameters at the last check-up between two groups. HCM-related death was defined as death caused by either SCD, congestive heart failure (CHF) or AF-related stroke (20). SCD was defined as instant and death unexpected within 1 h after a witnessing collapse in patients who previously were in a stable clinical condition, or nocturnal death with no antecedent history of worsening symptoms (21). Death caused by CHF was defined as death that occurred in context of progressive cardiac decompensation due to development of pulmonary edema or cardiogenic shock (21).

Statistical Analysis

All statistical analyses were done with SPSS 25.0 (IBM, Armonk, NY, United Status) and GraphPad Prism 8.0 (GraphPad Software Inc., La Jolla, CA, United States). Normally distributed measurement data are expressed as mean ± SD and non-normally distributed continuous data as median [interquartile range (IQR)]. In order to compare continuous variables, two independent sample *t*-test was used between two groups, while paired *t*-test was used within the same group. The chi-square or Fisher's exact test was used to compare non-continuous variables expressed as numerals (percentages). The Kaplan–Meier method with log-rank test was used to determine and compare the cumulative survival of different groups. To identify the prognostic predictors of all-cause mortality and HCM-related mortality, Cox regression model was used. First, the potential variables may affect all-cause mortality and HCM-related mortality were evaluated in a univariable model. Second, input variables with $P < 0.10$ into the backward stepwise multivariate analysis. It was considered statistically significant if *P*-values (2-sided) were less than 0.05.

RESULTS

Baseline Characteristics

Table 1 lists the baseline characteristics of 184 patients. Of these 184 patients, 102 were treated by medication (e.g., beta-receptor antagonists, calcium channel blockers) and 82 underwent SM. Patients in the SM group were younger (48.9 ± 10.4 years) than those in the MT group (55.1 ± 14.8 years, $p = 0.001$). Compared with MT group, the rate of comorbidity (particularly coronary artery disease and hypertension) of SM group was lower (26.8% vs. 52.9%, $p < 0.001$). LA diameter of the SM group was larger than that of the MT group (42.7 ± 7.3 vs. 39.5 ± 6.1 mm, $p = 0.001$). LVOT gradient at rest of SM group was 89.1 ± 35.7 mmHg, and it was obviously larger than that of MT group (66.3 ± 35.0 mmHg, $p < 0.001$).

TABLE 1 | Characteristics of 184 mildly symptomatic patients with HOCM at baseline.

Variable	MT (*n* = 102)	SM (*n* = 82)	*P*-value
Age (yrs)	55.1 ± 14.8	48.9 ± 10.4	0.001
Female (n,%)	43.0 (42.2)	32.0 (38.6)	0.667
BMI (kg/m^2)	25.8 ± 3.8	24.9 ± 3.4	0.128
SBP (mmHg)	123.7 ± 16.0	123.0 ± 15.8	0.797
DBP (mmHg)	74.4 ± 10.5	72.9 ± 10.7	0.339
Comorbidity (n,%)	54.0 (52.9)	22.0 (26.8)	<0.001
Coronary artery disease	18.0 (17.6)	5.0 (6.1)	0.019
Hypertension	41.0 (40.2)	17.0 (20.7)	0.005
Diabetes	7.0 (6.9)	2.0 (2.4)	0.299
HCM family history (n,%)	7.0 (6.9)	5.0 (6.1)	0.834
History of HOCM (yrs)	3.3 ± 4.5	3.7 ± 3.8	0.593
History of AF (n,%)	6.0 (5.9)	8.0 (9.8)	0.325
NYHA class	2.0 ± 0.0	2.0 ± 0.0	–
LA diameter (mm)	39.5 ± 6.1	42.7 ± 7.3	0.001
LV end-diastolic diameter (mm)	43.0 ± 5.0	43.6 ± 5.1	0.388
LV ejection fraction (%)	69.4 ± 7.5	69.0 ± 6.8	0.714
Septal thickness (mm)	20.5 ± 5.0	21.0 ± 5.3	0.593
Resting LVOT gradient (mmHg)	66.3 ± 35.0	89.1 ± 35.7	<0.001

MT, medical therapy; SM, septal myectomy; BMI, body mass index; SBP, systolic blood pressure; DBP, diastolic blood pressure; HCM, hypertrophic cardiomyopathy; HOCM, hypertrophic obstructive cardiomyopathy; AF, atrial fibrillation; NYHA, New York Heart Association; LA, left atrium; LV, left ventricular; LVOT, left ventricular outflow tract.

Procedure Data

A total of 82 patients underwent SM. There were five patients (5/82, 6.1%) died during the perioperative period in the SM group: four of them died of CHF, and one died of cerebral hemorrhage. In the perioperative period, one patient (1/82, 1.2%) implanted a permanent pacemaker due to third-degree atrioventricular block after SM.

Survival

Follow-up was completed in 179 patients and the median follow-up time was 5.0 years (IQR: 4.0 to 8.0 years, maximum: 18.0 years). In the period of follow-up, no patients accepted septal reduction therapy or implanted cardioverter defibrillator, either in the MT or SM groups. There were 14 deaths in the entire cohort during follow-up, including 11 deaths in the MT group (annual mortality rate: 0.6%/year) and 3 deaths in the SM group (annual mortality rate: 0.3%/year). The clinical endpoints of patients are summarized in **Table 2**. Cardiovascular death accounted for larger percentage (10/14, 71.4%) of all-cause death in this study, including 9 (9/11, 81.8%) in the MT group and 1 (1/3, 33.3%) in the SM group. There were 9 (9/102, 8.8%) patients died of HCM-related death in the MT group: 5 (5/102, 4.9%) due to SCD, 1 (1/102, 1.0%) due to CHF and 3 (3/102, 2.9%) due to AF-related stroke. However, in the SM group, only 1 (1/77, 1.3%) died of HCM-related death during the long-term follow-up, and the remaining 2 patients died of severe pneumonia (5 years after SM) and malignant tumors (6 years after SM), respectively. 5-year and 10-year overall survival of the SM group was 96.5% (95%CI: 91.6% to 100.0%) and 93.1% (95%CI: 84.9% to 100.0%), respectively. This survival was comparable to that of the MT

TABLE 2 | The classification of clinical endpoints during follow-up (n,%).

Variable	MT (*n* = 102)	SM (*n* = 77)	*P*-value
All-cause death	11 (10.8)	3 (3.9)	0.089
Cardiovascular death	9 (8.8)	1 (1.3)	0.066
Non-cardiovascular death	2 (2.0)	2 (2.6)	1.000
5-year overall survival	92.9%	96.5%	–
10-year overall survival	83.0%	93.1%	–
HCM-related death	9 (8.8)	1 (1.3)	0.066
SCD	5 (4.9)	0	–
CHF	1 (1.0)	1 (1.3)	1.000
AF-related stroke	3 (2.9)	0	–
5-year HCM-related survival	94.2%	98.7%	–
10-year HCM-related survival	86.1%	98.7%	–

MT, medical therapy; SM, septal myectomy; HCM, hypertrophic cardiomyopathy; SCD, sudden cardiac death; CHF, congestive heart failure; AF, atrial fibrillation.

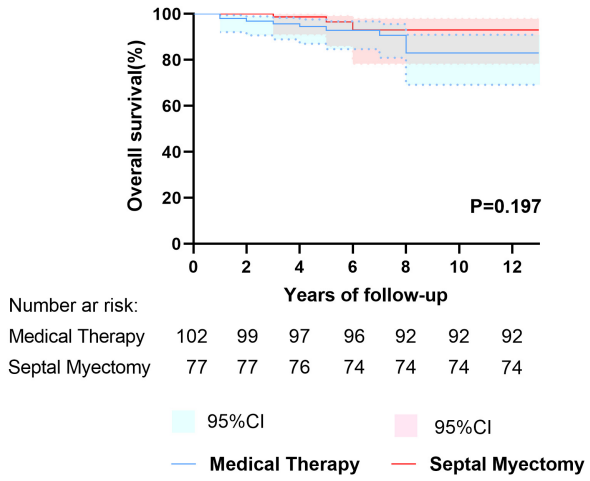

FIGURE 1 | Kaplan–Meier curves depicting overall survival between the septal myectomy and medical therapy groups.

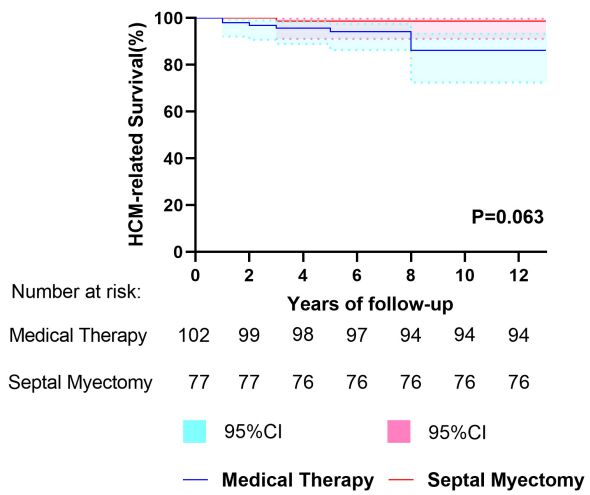

FIGURE 2 | Kaplan–Meier curves depicting HCM-related survival between the septal myectomy and medical therapy groups.

group, whose 5- and 10-year overall survival were 92.9% (95%CI: 87.4% to 98.4%) and 83.0% (95%CI: 72.4% to 93.6%), respectively (P = 0.197) (**Figure 1**). 5- and 10-year HCM-related survival for two groups was 98.7% (95%CI: 96.2% to 100.0%) and 98.7% (95%CI: 96.2% to 100.0%) vs. 94.2% (95%CI: 89.1% to 99.3%) and 86.1% (95%CI: 76.3% to 95.9%), respectively (P = 0.063) (**Figure 2**). Cox multivariate regression analysis suggested that resting LVOT gradient in the last clinical examination was an independent predictor of all-cause mortality (HR = 1.017, 95%CI: 1.000–1.034, P = 0.045) and HCM-related mortality (HR = 1.024, 95%CI: 1.005–1.043, P = 0.012) (**Table 3** and **Table 4**).

Clinical Outcome

Table 5 lists the clinical results of 179 patients. In the SM group, the clinical symptoms were remarkably improved (NYHA class post-SM: 1.3 ± 0.6, P < 0.001), and 56 patients (56/77, 72.7%) were in NYHA class I. Nevertheless, the patients' clinical symptoms did not improve after MT (NYHA class after medical treatment: 2.1 ± 0.5, P = 0.127). Resting LVOT gradient, with an average decrease of 78.5%, had reduced from 89.4 ± 35.5 to 16.7 ± 12.2 mmHg (p < 0.001) after SM. Moreover, there were 67 patients (67/77, 87.0%) after SM with a resting LVOT gradient < 30 mmHg. Meanwhile, the resting LVOT gradient of MT group, with an average decrease of 28.3%, was reduced from 66.3 ± 35.0 to 56.5 ± 27.7 mmHg (P = 0.001). However, there were only 22 patients (22/102, 21.6%) in the MT group with a resting LVOT gradient < 30 mmHg. Patients of the SM group had LA diameter reducing from 42.7 ± 7.3 to 37.7 ± 5.2 mm (P < 0.001). Instead, patients of MT group had left atrium diameter increasing from 39.5 ± 6.1 to 43.2 ± 6.9 mm (P < 0.001). During the period of following up, two patients (2/77, 2.6%) implanted permanent pacemaker due to third-degree atrioventricular block after SM.

TABLE 3 | Predictors of all-cause mortality.

Variable	Univariate		Multivariate	
	HR (95%CI)	*P*-value	HR (95%CI)	*P*-value
Baseline				
Age (yrs)	1.035 (0.991–1.082)	0.120	–	–
Female	1.380 (0.463–4.112)	0.563	–	–
History of AF (n,%)	2.247 (0.496–10.175)	0.293		
SM	0.438 (0.120–1.602)	0.212	–	–
LA diameter (mm)	1.002 (0.926–1.085)	0.952	–	–
LV end-diastolic diameter (mm)	0.952 (0.858–1.056)	0.350	–	–
LV ejection fraction (%)	0.980 (0.909–1.058)	0.609	–	–
Septal thickness (mm)	1.006 (0.893–1.132)	0.924	–	–
Resting LVOT gradient (mmHg)	0.999 (0.984–1.132)	0.929	–	–
Follow up				
New-onset AF	0.729 (0.161–3.293)	0.670	–	–
LA diameter (mm)	1.040 (0.967–1.120)	0.291	–	–
LV end-diastolic diameter (mm)	0.911 (0.816–1.017)	0.097	0.942 (0.836–1.062)	0.331
LV ejection fraction (%)	1.081 (1.000–1.168)	0.049	1.050 (0.969–1.138)	0.230
Septal thickness (mm)	1.052 (0.932–1.186)	0.414	–	–
Resting LVOT gradient (mmHg)	1.020 (1.003–1.036)	0.022	1.017 (1.000–1.034)	0.045

HR, hazard ratio; CI, confidence interval; AF, atrial fibrillation; SM, septal myectomy; LA, left atrium; LV, left ventricular; LVOT, left ventricular outflow tract.

DISCUSSION

This study firstly and directly compares the outcome of SM and MT in mildly symptomatic patients with HOCM. The crucial findings of this study were listed as follows: (1) overall survival and HCM-related survival of SM group were comparable

TABLE 4 | Predictors of HCM-related mortality.

Variable	Univariate		Multivariate	
	HR (95%CI)	*P*-value	HR (95%CI)	*P*-value
Baseline				
Age (yrs)	1.041 (0.998–1.098)	0.132	–	–
Female	2.064 (0.554–7.700)	0.280	–	–
History of AF (n,%)	3.720 (0.770–17.975)	0.102		
SM	0.175 (0.022–1.411)	0.102		–
LA diameter (mm)	0.982 (0.893–1.080)	0.710	–	–
LV end-diastolic diameter (mm)	0.965 (0.851–1.093)	0.575	–	–
LV ejection fraction (%)	0.969 (0.885–1.060)	0.488	–	–
Septal thickness (mm)	1.006 (0.878–1.153)	0.931	–	–
Resting LVOT gradient (mmHg)	1.001 (0.983–1.019)	0.952	–	–
Follow up				
New-onset AF	1.136 (0.235–5.480)	0.874	–	–
LA diameter (mm)	1.043 (0.956–1.138)	0.347	–	–
LV end-diastolic diameter (mm)	0.943 (0.828–1.073)	0.372	–	
LV ejection fraction (%)	1.097 (0.996–1.209)	0.059	1.079 (0.982–1.185)	0.113
Septal thickness (mm)	1.086 (0.969–1.217)	0.156	–	–
Resting LVOT gradient (mmHg)	1.027 (1.007–1.046)	0.007	1.024 (1.005–1.043)	0.012

HR, hazard ratio; CI, confidence interval; AF, atrial fibrillation; SM, septal myectomy; LA, left atrium; LV, left ventricular; LVOT, left ventricular outflow tract.

TABLE 5 | Clinical and echocardiographic characteristics of 179 mildly symptomatic patients with HOCM at the last check-up.

Variable	MT (*n* = 102)	SM (*n* = 77)	*P*-value
NYHA class	2.1 ± 0.5	1.3 ± 0.6[b]	< 0.001
NYHA class I (n,%)	7.0 (6.0)	56.0 (72.7)	< 0.001
NYHA class II (n,%)	81.0 (79.4)	17.0 (22.1)	< 0.001
NYHA class III (n,%)	14.0 (13.7)	3.0 (3.9)	0.026
NYHA class IV (n,%)	0.0 (0.0)	1.0 (1.3)	0.430
NYHA class III/IV (n,%)	14.0 (13.7)	4.0 (5.2)	0.060
New-onset AF (n,%)	20 (20.8)	13 (18.8)	0.752
LA diameter (mm)	43.2 ± 6.9[b]	37.7 ± 5.2[b]	< 0.001
LVend-diastolic diameter (mm)	43.8 ± 4.9	42.9 ± 5.4	0.238
LV ejection fraction (%)	66.2 ± 7.2[b]	62.7 ± 6.8[b]	0.001
Septal thickness (mm)	19.9 ± 4.4	17.8 ± 4.6[b]	0.002
Resting LVOT gradient (mmHg)	56.5 ± 27.7[a]	16.7 ± 12.2[b]	< 0.001
Reduction in LVOT gradient (%)	28.3 ± 18.4	78.5 ± 18.6	< 0.001

MT, medical therapy; SM, septal myectomy; NYHA, New York Heart Association; AF, atrial fibrillation; LA, left atrium; LV, left ventricular; LVOT, left ventricular outflow tract; [a]*P < 0.01 and* [b]*P < 0.001 compared with the baseline characteristics.*

to those of MT group; (2) compared to the MT, SM had advantages on improving clinical symptoms and reducing resting LVOT gradient; (3) resting LVOT gradient at the last clinical check-up was an independent predictor of all-cause mortality and HCM-related mortality in mildly symptomatic patients with HOCM.

Now, SM has been proven by multiple previous studies that exerts a positive effect on long-term prognosis for HOCM patients with severe symptoms (7, 8, 22). It is worth noting that a recent study conducted by Desai and his colleagues concerning a large proportion of mildly symptomatic or asymptomatic patients (88% patients with NYHA class I/II) with HOCM demonstrated that the composite event (death except non-cardiac causes and/or appropriate ICD discharge) rate of MT group was twice as high as that of SM group (76% patients with NYHA class I/II) (23). However, compared with MT, whether early surgery could provide better survival for mildly symptomatic patients with HOCM is not yet known. Therefore, the present study was dedicated to discuss this issue.

In our study, 10-year overall survival of patients in MT group was 83.0%. Similar to our data, a study conducted by Vriesendorp et al. (24) reported a 10-year overall survival of 84.0% of MT group, but their study cohort were mildly symptomatic or asymptomatic (NYHA class I/II) patients with HOCM. Our survival rate of MT group was higher compared with two other studies with 10-year overall survival of 75.8% (Ball et al., 33.3% patients of MT group in NYHA class III/IV) (25) and 72.2% (Yin-Jian Yang et al., 44.4% patients of MT group in NYHA class III/IV) (26). This might be due to the fact that these two studies involved some HOCM patients with NYHA class III/IV in the MT group, and multiple studies have demonstrated that for patients with HOCM, NYHA class III/IV is independently associated with worse prognosis (8, 27). In the present study, mildly symptomatic patients with HOCM after SM had comparable overall survival and HCM-related survival to those treated with medication. But compared with MT, SM had obvious advantages on maintaining long-lasting improvement in symptoms. Our data indicated that the clinical symptoms was remarkably improved after SM (NYHA class post-SM: 1.3 ± 0.6, $P < 0.001$), and 56 patients (72.7%) were in NYHA class I. However, the patients' clinical symptoms did not improve after MT (NYHA class after medical treatment: 2.1 ± 0.5, $P = 0.127$). Furthermore, our data indicated that 10-year overall survival and HCM-related survival of SM group were 93.1% and 98.7%, respectively, which evidently higher than those reported by Ball et al. (25) concerning HOCM patients some with severe symptoms (33.3% patients with NYHA class III/IV) in the MT group (10-year overall survival and HCM-related survival 75.8 and 86.9%, respectively). Recently, outcome of earlier surgery vs. surgery for guideline-based Class I indication in patients with HOCM was discussed by Alashi et al. (13). In their study, earlier surgery was applied to patients who were in NYHA class II with drug intolerance or who were in NYHA class I but with symptomatic impairment of exercise capacity despite optimal medical therapy. The data of Alashi et al. (13) indicated that for patients with HOCM, earlier surgery vs. surgery for Class I indication was associated with a higher long-term survival, close to the age- and sex- matched US population. Therefore, combined the above important findings, we considered that

earlier surgical intervention may be a reasonable option for mildly symptomatic patients with HOCM who intolerant to MT, rather than only undergoing watchful waiting.

Multiple studies have demonstrated that the prognosis of HCM patients with obstruction is poorer than that of those without obstruction, especially for patients with severe symptoms (7, 28, 29). Moreover, a study conducted by Sorajja et al. (11) suggested that for mildly symptomatic or asymptomatic HOCM patients, an elevated LVOT gradient was independently associated with higher risk of developing heart failure and death. Similarly, results of our study suggested that resting LVOT gradient in the last clinical examination was an independent predictor of all-cause mortality in mildly symptomatic patients with HOCM, and every 1 mmHg increase added the risk of all-cause mortality by 1.7%. Additionally, our data suggested that resting LVOT gradient in the last clinical examination was also an independent predictor of HCM-related mortality, and every 1 mmHg increase added the risk of HCM-related mortality by 2.4%. Additionally, in multiple clinical trials, SM has been proven to be able to safely and effectively reduce the LVOT gradient in patients with HCM (8, 30, 31). Likewise, our research results also confirmed this point. Moreover, the present study indicated that compared with MT, SM had advantages on reducing resting LVOT gradient (average decrease on resting LVOT gradient: 78.5% vs. 28.3%, $P < 0.001$). Consequently, in order to reduce the negative impact of high LVOT gradient, SM is also seemed to be a reasonable choice for mildly symptomatic patients with high LVOT gradient who intolerant to drug treatments.

There are several limitations in this study. First, this was a retrospective study with a small sample from a single-center, a relatively experienced HCM management center in China. Therefore, our results were limited by referral and selection bias and might not be generalizable to else centers. Second, in this study, level of symptoms of patients was based on self-statement. However, some patients may adapt themselves to their restriction of exercise capacity and thus report a lower degree of symptom severity, which may influence the baseline characteristics and clinical results of follow-up in this study. Third, an advantage of this research was that we compared the prognosis of conservative treatment and surgical intervention, but overall survival and HCM-related survival of patients enrolled in this research did not make a comparison with expected survival of an age- and sex-matched Chinese general population.

CONCLUSION

Compared with MT, SM had comparable overall survival and HCM-related survival in mildly symptomatic HOCM patients, but SM had advantages on improving clinical symptoms and reducing resting LVOT gradient. Resting LVOT gradient in the last clinical examination was an independent predictor of all-cause mortality and HCM-related mortality.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Medicine Ethics Committee of Beijing Anzhen Hospital. Written informed consent for participation was not required for this study in accordance with the national legislation and the institutional requirements.

AUTHOR CONTRIBUTIONS

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

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CASE REPORT
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Case Report: Double-Decker Repair of Partial Pulmonary Venous Return Into the Coronary Sinus

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We present a case of persistent left superior vena cava (LSVC) draining into the right atrium (RA) *via* the coronary sinus (CS), while the left superior pulmonary vein returns abnormally to the CS. The LSVC may have few clinical consequences but complicates surgical repair of partial anomalous pulmonary venous return (PAPVR). Transthoracic echocardiography and computed tomographic angiography (CTA) showed that a persistent LSVC and PAPVR converged behind the left atrium. During the operation, the left atrium was adjacent to the confluence part. We resected a portion of the adjacent left atrium to create an inlet of the pulmonary veins and used two autologous pericardial patches to reconstruct a tunnel directing flow from the left pulmonary veins to the surgically created inlet in the adjacent left atrium, and another upper tunnel directing flow from the LSVC to the dilated CS. Pulmonary CTA confirmed that both PAPVR flow to LA and LSVC flow to RA were unobstructed. At a 12-month follow-up, the patient was asymptomatic. No supraventricular arrhythmia was detected. We would like to present this additional technique to our armamentarium to treat PAPVR in combination with LSVC.

Keywords: partial anomalous pulmonary venous return, persistent left superior vena cava, congenital heart disease, cardiac surgery, pulmonary vein

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INTRODUCTION

Partial anomalous pulmonary venous return (PAPVR) is the most common type of pulmonary venous return anomaly (1). The surgical procedure for PAPVR drainage into the coronary sinus (CS) is controversial, especially in conjunction with a left superior vena cava (LSVC) draining to an intact CS. The combination of these two congenital defects was managed by using two patches to restore the LSVC to the CS in combination with the restoration of normal connection of pulmonary venous drainage to the left atrium in this case report.

CASE DESCRIPTION

A 27-year-old female patient presented to our institution due to palpitations for 2 months. The patient has no history of shortness of breath or fatigue. She gave birth to a child 5 months ago. During pregnancy, her heart palpitations were very severe, and diuretics could partially relieve her. At the initial visit to cardiac surgery, her height was 164 cm, and weight was 60 kg. Her blood pressure, heart rate, and respiratory were normal. This patient has no murmur.

Transthoracic echocardiography showed a persistent LSVC draining into the CS and anomalous left pulmonary veins draining into the intact and dilated CS. LSVC and PAPVR converged behind the left atrium. Bilateral superior vena cava of comparable size can be seen. We could not detect the innominate vein. The right heart structure was dilated. Mimics software (20.0, Materialise, Belgium) was used to reconstruct the 3-dimension of congenital heart disease (**Figure 1**). According to the 3-D model, the left atrium is adjacent to the confluence of LSVC and PAPVR.

The patient underwent surgical repair. Standard median sternotomy was performed. Cardiopulmonary bypass was established using standard ascending aortic and bicaval cannulation. After the heart was arrested, a persistent LSVC cannulation was inserted through the enlarged CS. The confluence of LSVC and PAPVR was explored and detected. The left atrium was adjacent to the confluence part. We resected the partial adjacent left atrium (about 2*3 cm) to create an inlet of pulmonary veins. A surgically created tunnel was formed from a baffle of the autologous pericardium that is sewn inside the left pulmonary vein in such a way as to direct flow from the left pulmonary veins to the surgically created inlet in the adjacent left atrium. The LSVC was left there. Then, we used another pericardium to create another upper tunnel to direct flow from the LSVC to the dilated CS (see **Supplemental Material** and **Figure 2**), which overrode the reconstructed pulmonary vein. The reconstruction tunnel that leads the left pulmonary veins to the left atrium is located below the new tunnel that leads the LSVC to the CS. The inferior pericardium was shared as part of the ventral wall of the LSVC channel and the dorsal wall of the pulmonary venous channel. We used 6/0 Prolene on the 8-mm needle to suture the natural pericardial patch and took care to avoid the narrowing of the two tunnels during the operation. Pulmonary CTA confirmed that both PAPVR flow to LA and LSVC flow to RA were unobstructed (see **Figure 1**). The pulmonary venous drainage was widely patent. At a 12-month follow-up, the patient was asymptomatic. No supraventricular arrhythmia was detected.

DISCUSSION

PAPVR represents a physiological left-to-right shunt, which may increase the subsequent risk for pulmonary vascular disease, biventricular failure, and Eisenmenger syndrome (2). Surgical correction of PAPVR is commonly carried out using internal patch technology or its modification and the Warden

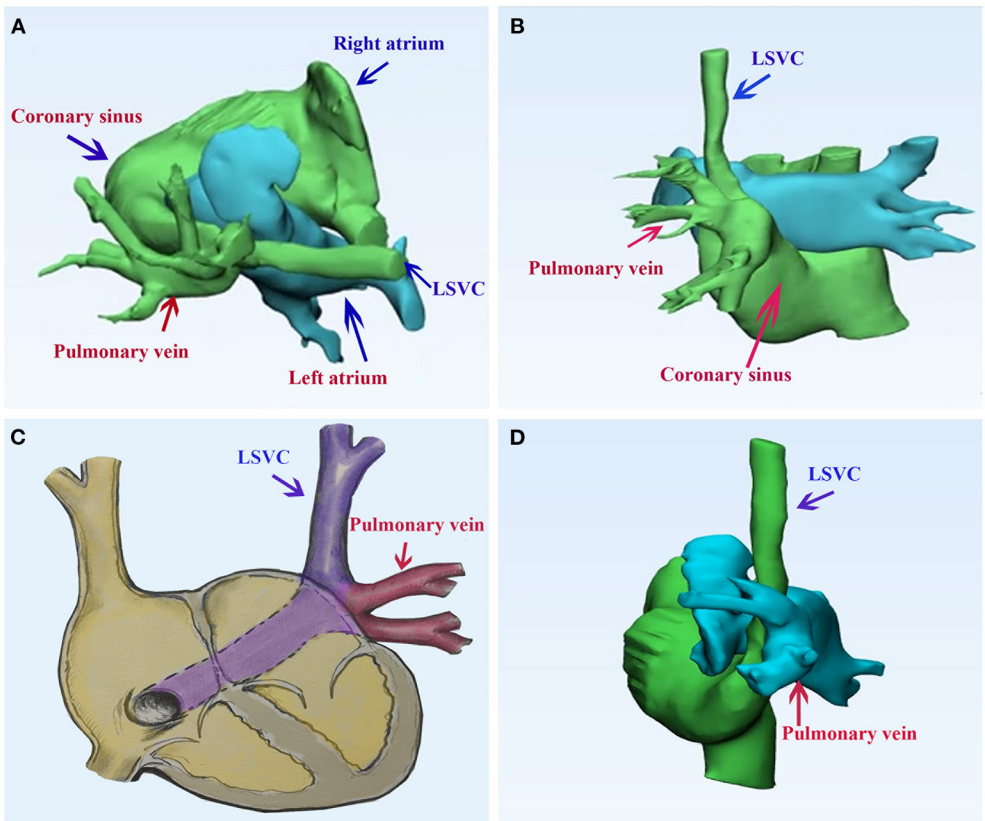

FIGURE 1 | Pulmonary computed tomography of the case of persistent left superior vena cava draining into the right atrium *via* the coronary sinus, while the left superior pulmonary vein returns abnormally to the coronary sinus. **(A,B)** The 3-dimension model of the congenital heart disease; **(C)** a diagram of the case; **(D)** the postoperative 3-dimension model of the double-decker technique. LSVC, left superior vena cava.

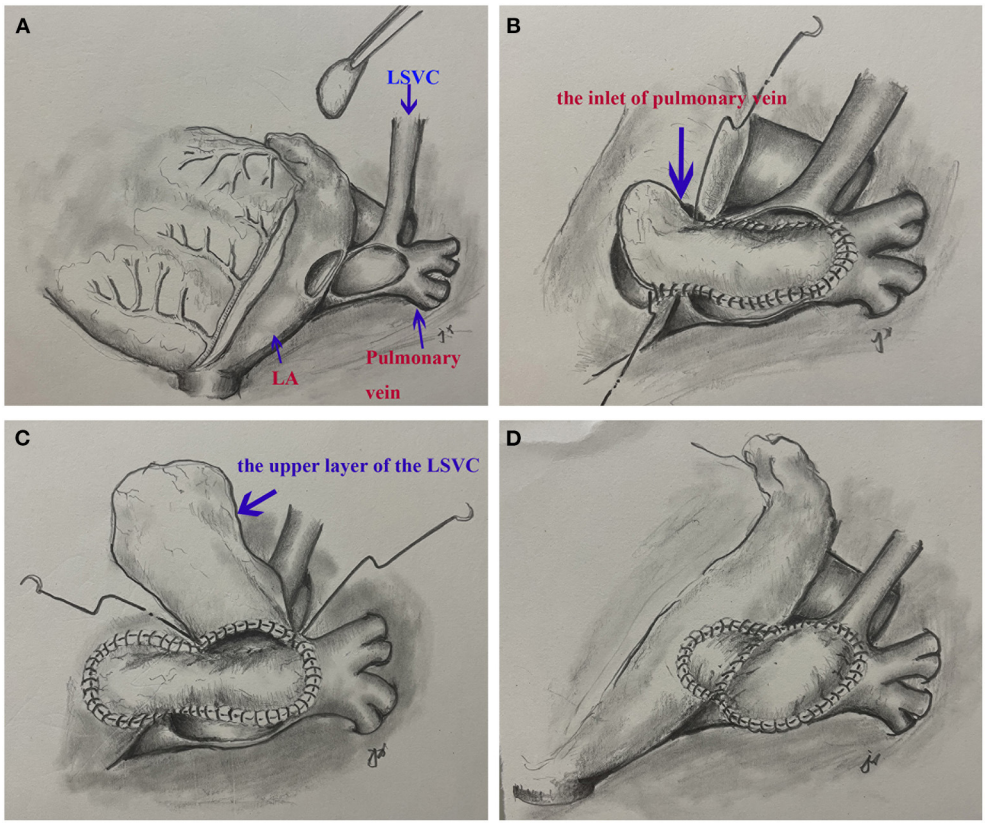

FIGURE 2 | A surgical view of the double-decker technique. **(A)** Resection of a portion of left atrium and cut the confluence of the left superior vena cava and pulmonary veins. **(B)** The autologous pericardial baffle is sutured by guiding the blood flow of the left pulmonary vein to the surgical entrance of the adjacent left atrium. **(C)** The other pericardium creates another upper tunnel, directing blood flow from the left superior vena cava to the dilated coronary sinus. **(D)** The completed double-decker technique.

procedure (4). By baffling these structures with pericardium or a bovine patch, performing or without patch cavity angioplasty as needed, thereby redirecting abnormal pulmonary venous return through the intra-atrial defect. However, when there is no atrial defect and LSVC is present, the best surgery for a complete repair cannot be determined.

The incidence of persistent LSVC is ∼0.3%, and the incidence of PAPVR is 0.6–0.7% (3). We can only identify one report on this rare combination of systemic and pulmonary venous anomalies, which together drainage into the CS (3). This combination complicates the repair of PAPVR. Ligating the LSVC may be one choice if tolerated; we had carefully measured the diameter of the LSVC and examined the CTA before the procedure. We found the LSVC was similar in size to the right superior vena cava, and there was no decompressing bridging vein. Ligation of the LSVC may lead to venous congestion and possible intracerebral hemorrhage if there is no bridging vein or the vein is of insufficient size. Extracardiac techniques involving transposition of the LSVC to the right atrial appendage and unroofing of the CS to the LA are an alternative method, just like the modified Warden procedure on the right side. The Warden procedure was used to deal with multiple-level entry pulmonary veins, without transecting or manipulating the cavoatrial junction. For right-sided PAPVR, this approach reduces the incidence of sinus rhythm loss but carries the risk of SVC obstruction. Insufficient length of LSVC is a critical limitation of adult extracardiac technology. We measured the length of the LSVC and found that the LSVC was not long enough to connect directly to the right atrial appendage. Furthermore, we have to perform an overall dissection of LSVC, which is somewhat complex and time-consuming compared to our technique. Also, the deoxygenated coronary venous return to the LA was inevitable *via* intra-atrial baffling. Another alternative method is to tunnel the LSVC to the right on the roof of the atrium. It can also achieve the surgical treatment aim of PAPVC draining into the right atrium: complete correction by rerouting the abnormal pulmonary veins to the left atrium and redirecting the LSVC to the right atrium. Due to the relatively large size of LSVC, the tunnel should be created carefully without obstruction. By using our method, there is no residual abnormal systemic drainage, and no intra-atrial manipulation is required. An alternative approach is to isolate the left pulmonary vein (with the resulting defect patch) followed by anastomosis with the left atrium/left atrial appendage, which seems simpler. However, we found insufficient tissue for direct

anastomosis. Direct anastomosis may also lead to subsequent pulmonary vein stenosis and LSVC compression. We also avoid the end-to-end anastomosis between the LSVC and the RAA and decrease the risk of late stenosis of the systemic venous chamber.

To facilitate unobstructed LSVC drainage and right pulmonary vein drainage is critical. For drainage of the right pulmonary vein, we preserved the natural venous tissue underneath to preserve the growth potential and prepared the appropriate pericardial patch to create a tension-free tunnel and prevent late pulmonary vein stenosis. Nevertheless, pulmonary venous stenosis may develop due to the shrinkage of an untreated autologous pericardial patch used to baffle the venous drainage (4). It still needs further exploration.

Double-decker repair is introduced by Hongu (5). The author developed a new surgical technique with minimum right atriotomy and double-barreled arrangement of systemic and pulmonary venous channels. In his report, pulmonary venous blood flows through the proximal SVC, intra-atrial tunnel, and venous sinus defect into the left atrium. Systemic venous blood flows through the ventral opening of the SVC and the RAA chicane that runs across the proximal SVC into the RA. He also created a surgical ASD in patients with an intact atrial septum. Our method is similar to his previous report. But due to the different anatomical entities, our method is easier to understand. The direction of the reconstructed tunnels is parallel to the original vessels. We speculate that our technique would reduce turbulence and achieve good long-term results.

Supraventricular arrhythmia following surgical repair is an important and distressing feature (4). But most of it occurs in patients who have incisions or suturing through the anterior right atrial-superior vena cava junction, which can lead to the risk of sinus node dysfunction. For our technology, we speculate that any type of arrhythmia is less likely to occur. In addition, postoperative anticoagulation is not required.

After all, the advantages of our technique are numerous: (1) the integrity of the left atrium, right atrium, and atrial septum was preserved; (2) both tunnels retained growth potential; (3) anastomosis and manipulation of the entire circumference of the LSVC and PAPVR are avoided; (4) there is no residual anomalous pulmonary or systemic drainage. The Warden, single-patch, double-patch, and double-decker techniques (5) are complementary options for the treatment of PAPVR, and the choice depends on the location of the sinus node, the extent of the right atrium, the location of the pulmonary vein, the size of SVC, and so on.

DATA AVAILABILITY STATEMENT

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

ETHICS STATEMENT

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

AUTHOR CONTRIBUTIONS

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

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fcvm.2022.853005/full#supplementary-material

Supplementary Video 1 | The procedure of the double-decker technique.

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ORIGINAL RESEARCH
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Correlation Between Post-operative Sense of Coherence and Family Function in Patients With Type A Aortic Dissection

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Objective: To analyze the relationship between post-operative sense of coherence and family function in patients with type A aortic dissection (AD).

Methods: Ninety patients with AD treated from January 2019 to December 2020 were selected as the research subjects. All patients received surgical treatments. Two weeks after the operation, the Sense of Coherence Scale (SOC-13) and Family APGAR index scale (APGAR) were used to evaluate the patients' sense of coherence and family function. Baseline data of all patients were collected, the SOC-13 scores of patients with type A AD with different demographic characteristics were compared, and the relationship between family function and patients' sense of coherence was analyzed.

Results: The ninety patients with type A AD had a low level of psychological consistency, and the average SOC-13 score was 49.84 ± 3.89 points. The SOC-13 score of patients with type A AD with family monthly incomes <5,000 yuan and moderate and severe family dysfunction was lower than that of patients with family monthly incomes ≥5,000 yuan and good family function. The difference was statistically significant ($P < 0.05$). There was no statistically significant difference in the SOC-13 scores of patients with type A AD with different demographic characteristics ($P > 0.05$). The results of multiple linear regression analysis showed that family monthly income <5,000 yuan and moderate and severe family dysfunction might be general influencing factors of sense of coherence among patients with type A AD ($P < 0.05$). $Y = 43.333 + 6.667X_1 + 16.730X_2$ was obtained.

Conclusion: The post-operative sense of coherence of patients with type A AD may be affected by family function.

Keywords: type A aortic dissection, sense of coherence, family function, correlation, research

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INTRODUCTION

As the preferred treatment for patients with type A aortic dissection (AD), surgical treatment aims to prevent aortic rupture and cardiac tamponade, improve patient hemodynamics and reduce mortality in patients with type A AD (1). Due to the serious condition of type A AD, the patient may have persistent pain and need lifelong antihypertensive therapy after the operation (2, 3). Psychological congruence is the individual's perception of life as a whole, and it can reflect the

individual's internal psychological tendency (4). Research indicates that the level of psychological congruence can influence the self-management ability of patients and improve their quality of life (5). Studies in China and internationally have shown that (6–9) among post-operative AD patients, a psychological status oriented toward improving quality of life, especially for long-term (6 months or more) quality of life, is positively related to a good psychological state; patients with a better psychological status have a better understanding of stressors and a better physical status, actively engage in health-related behavior, have better self-management ability, and thus have a better quality of life. Research by Fanghui Lou (10) and Zhao Rong (11) showed that psychological intervention for patients after AD can correct negative emotions, improve psychological status, promote patients' sense of psychological consistency and improve their quality of life. Therefore, it is important to clarify the status quo of psychological congruence in patients with type A AD and to determine the related factors for improving their quality of life. Family function mainly refers to the family's ability to satisfy various psychological and physiological development needs of family members (12). Studies have shown that good family functioning contributes to patients' self-management ability (13). Combined with the influence of the abovementioned psychological congruence and family function on the self-management ability of patients with type A AD, it is inferred that there may be some relationship between psychological congruence and family function. In view of this, this study focuses on a correlation analysis of type A AD with post-operative psychological coherence and family function. The report is as follows.

MATERIALS AND METHODS

General Data

Ninety patients with AD admitted from January 2019 to December 2020 were selected as subjects for this study, which was approved by the hospital ethics committee. All patients and their families were informed about the study and voluntarily signed the informed consent form. Among the 90 patients, 64 were male, and 26 were female. The average age was 40–75 years (57.82 ± 3.58 years). Regarding education level, 28 patients were educated at the junior high school level or below, 39 patients were educated at the senior high school level or technical secondary school, and 23 patients were educated at the junior college level or above.

Inclusion and Exclusion Criteria

The Inclusion Criteria Were as Follows

① Patients had AD that met the relevant standards in internal medicine (9th Edition) (14) and was diagnosed as Stanford type A AD by echocardiography and enhanced vascular CT; ② patients had surgical treatments, including the Bentall operation, Wheat operation or ascending aorta replacement, that were performed for the first time; and ③ patients had good communication and comprehension skills and were able to complete the questionnaire independently.

The Exclusion Criteria Were as Follows

① Patients had complicated AD with serious complications, such as respiratory failure, cardiac respiratory arrest, renal failure, cerebral infarction, cerebral hemorrhage, coma, shock, or heart failure; ② patients had AD combined with malignant tumor diseases such as liver cancer or lung cancer; ③ patients had mental illness or cognitive impairment and were unable to cooperate with the investigation; or ④ patients withdrew or discontinued their participation halfway through the study.

Methods

Assessment of Psychological Consistency

Psychological consistency was evaluated 2 weeks after the operation, and the Sense of Coherence scale (SOC-13) (15) was used to evaluate the psychological consistency of patients. The scale includes 3 dimensions, including sense of control (items 3, 4, 5 and 10, with a total score of 4–28 points), sense of understanding (items 2, 6, 7, 8, and 12, with a total score of 5–35 points), and sense of meaning (items 1, 9, 11, and 13, with a total score of 4–28 points). Each item is scored from 1 to 7 points according to 7 levels, and the total score range is 13–91 points. The higher the score is, the higher the level of psychological consistency of patients. According to the scores, the level of patients' sense of psychological consistency was classified, with scores of 13–63 being classified as a low level, scores of 64–79 being classified as a medium level, and scores of 80–91 being classified as a high level. Patients selected each item in the questionnaire according to their own situations, and the score of each item ranged from 1 to 7 points. The researcher summed the scores for each item according to the patient's responses, and the final total score was the patient's sense of psychological consistency score (**Table 1**).

Family Function Assessment

Two weeks after the operation, the Family APGAR index (APGAR) (16) was used to assess the patient's family function. The scale consists of five dimensions: cooperation, emotion, adaptability, intimacy and growth. Each item is scored as 2, 1, or 0, which correspond to frequently, sometimes or rarely, respectively. The total score range is 0–10. The higher the score is, the better the family function. A total score $\leq$3 points indicates severe impairment of family function, 4–6 points indicates moderate impairment of family function, and $\geq$7 points indicates good family function. Patients rated each item of the scale according to their own situations, and the score of each item ranged from 0 to 2 points. The researcher summed the scores of the 5 items according to the patient's responses, and the final total score was the patient's family function score (**Table 2**).

Baseline Data Collection Design

A baseline data questionnaire was administered to collect baseline data of all patients, including age (<60 years old, $\geq$60 years old), sex (male and female), education level (junior middle school and below, senior high school or technical secondary school, college and above), marital status (married, unmarried/divorced/widowed), working status

TABLE 1 | Sense of coherence-13, SOC-13.

Here is a life orientation scale with seven different answers to each question, ranging from less to more severe. Please tick "√" according to your own feelings.

Item	Option	Score
1. Do you often feel like you don't care what's going on around you?	① never ② less ③ very little ④ hard to say ⑤ sometimes ⑥ more ⑦ often	
2. How often in the past have you been surprised by someone you thought you knew well?	① never ② less ③ very little ④ hard to say ⑤ sometimes ⑥ more ⑦ often	
3. How often has someone you count on let you down?	① never ② less ③ very little ④ hard to say ⑤ sometimes ⑥ more ⑦ often	
4. Do you often feel that you are being treated unfairly?	① never ② less ③ very little ④ hard to say ⑤ sometimes ⑥ more ⑦ often	
5. Do you often feel like you're in strange situations and don't know what to do?	① often ② sometimes ③ at individual times ④ I don't know ⑤ less ⑥ seldom ⑦ hardly or never	
6. Do you often have very complicated, mixed feelings and thoughts?	① very often ② often ③ sometimes ④ I don't know ⑤ less ⑥ seldom ⑦ hardly or never	
7. Do you often feel emotions you don't want to feel?	① very often ② often ③ sometimes ④ I don't know ⑤ less ⑥ seldom ⑦ hardly or never	
8. Many people, even very talented people, sometimes feel like failures under certain circumstances. Have you experienced this often in the past?	① never ② less ③ very few ④ hard to say ⑤ sometimes ⑥ more ⑦ often	
9. How often do you think that doing these things every day doesn't mean anything?	① very often ② often ③ sometimes ④ I don't know ⑤ less ⑥ seldom ⑦ hardly or never	
10. Do you often feel out of control?	① very often ② often ③ sometimes ④ I don't know ⑤ less ⑥ seldom ⑦ hardly or never	
11. Your life so far:	① no goals at all ② no goals ③ not very purposeful ④ unclear? have a life goal ⑤ clear life purpose ⑥ very clear life purpose	
12. When confronted with a problem or issue, you often find yourself—	① underestimating or overestimating its importance ② finding the matter hard to assess ③ addressing the matter slightly inaccurately ④ finding the matter hard to grasp ⑤ assessing the matter somewhat accurately ⑥ assessing the matter accurately ⑦ assessing the matter very accurately	
13. Doing the things you do every day—	① brings a great deal of happiness and satisfaction ② makes me happier and more satisfied ③ makes me a little happy ④ I don't know ⑤ makes me a little unhappy ⑥ makes me very unhappy? is a source of pain and trouble	
	Total points	

TABLE 2 | Family care index questionnaire (APGAR).

Item	Often (2 points)	Sometimes (1 point)	Hardly ever (0 point)
When I have problems, I can get satisfactory help from my family.	☐	☐	☐
I am very satisfied with the way my family discusses various things and shares problems with me.	☐	☐	☐
When I wish to pursue new activities or developments, my family is receptive and supportive.	☐	☐	☐
I am satisfied with the way my family shows concern and love for my emotions (joy, anger, sorrow, joy).	☐	☐	☐
I'm happy with the way my family spends time with me.	☐	☐	☐

(employed, retired), care (family members, others, no care), and family monthly income (<5,000 yuan, ≥5,000 yuan).

Quality Control

Before the study questionnaire was distributed, the purpose and reporting method of the survey was explained to the patients. The questionnaire was evenly distributed to the patients by two staff members to ensure that the patients completed the questionnaire independently within 30 min. After completion, the staff collected the questionnaire, checked whether the questionnaire was valid and eliminated invalid questionnaires (either all items were the same answer or there was a regular pattern of answers), included the valid questionnaires and entered the data. A total of 90 questionnaires were distributed, and 90 were recovered, for a recovery rate of 100%.

TABLE 3 | Comparison of the SOC-13 scores of patients with type A AD with different demographic characteristics.

Characteristics		*n*	SOC-13 score (points)	Statistical value	*P*
Age	<60 years old	68	49.77 ± 3.46	$t = 0.322$	0.748
	≥60 years old	22	50.06 ± 3.81		
Gender	Male	64	49.89 ± 2.98	$t = 0.281$	0.779
	Female	26	49.72 ± 3.16		
Education level	Junior high school and below	28	49.22 ± 2.95	$F = 0.641$	0.529
	High school or technical secondary school	39	50.14 ± 3.56		
	College degree or above	23	50.09 ± 3.99		
Marital status	married	69	49.52 ± 3.65	$t = 1.473$	0.144
	Unmarried/divorced/widowed	21	50.89 ± 3.58		
Working status	On the job	68	50.11 ± 3.91	$t = 1.171$	0.245
	Retired	22	49.01 ± 3.63		
Care situation	Family members	62	50.08 ± 3.09	$F = 0.831$	0.439
	Someone else	22	49.58 ± 4.17		
	Unattended	6	48.31 ± 3.25		
Monthly household income	<5,000 yuan	9	32.58 ± 2.70	$t = 11.383$	<0.001
	≥5,000 yuan	81	51.76 ± 4.96		
Family function	Good family function	63	60.21 ± 5.78	$F = 449.560$	<0.001
	Moderate impairment of family function	19	25.41 ± 2.09[a]		
	Severe family dysfunction	8	26.20 ± 2.25[a]		

Compared with family function, [a]the difference is statistically significant.

Statistical Methods

The data were processed by SPSS 22.0 software. The measurement data are expressed with respect to the normal distribution proposed by the Shapiro–Wilk normal distribution test. The independent sample *t*-test was used for intergroup comparisons, the one-way variance test was used for multigroup data, and the SNK test was used for pairwise comparisons. Multiple linear regression analysis was used to test the effect of family function on psychological consistency in patients with type A AD; $P < 0.05$ was considered statistically significant.

RESULTS

Post-operative SOC-13 Score of Patients With Type A AD

The ninety patients with type A AD had a low level of psychological consistency. The average SOC-13 score was 49.84 ± 3.89.

Comparison of SOC-13 Scores of Patients With Type A AD With Different Demographic Characteristics

The SOC-13 score of patients with type A AD with family monthly incomes <5,000 yuan and moderate and severe family dysfunction was lower than that of patients with family monthly incomes ≥5,000 yuan and good family function ($P < 0.05$). There was no significant difference in SOC-13 scores among patients with type A AD with different demographic characteristics ($P > 0.05$) (**Table 3**).

Linear Regression Analysis of the Influence of Various Factors on the Sense of Psychological Consistency of Patients With Type A AD

Taking the psychological consistency (SOC-13 score) of patients with type A AD as the dependent variable and the variable with a statistically significant difference of 2.2 as the independent variable, the equation was obtained by multiple linear regression analysis: $y = 43.333 + 6.667x1 + 16.730x2$, in which the F value of the regression model was 70.064, R2 was 0.617, and the adjusted R2 was 0.608. The results showed that family monthly income <5,000 yuan and moderate and severe family dysfunction may be influencing factors of general psychological consistency in patients with type A AD ($t = 2.301$, 8.822, all $P < 0.05$) (**Table 4**).

DISCUSSION

Affected by pain associated with the disease, surgical stress and disease management, patients with type A AD have poor psychological adaptability, low confidence in rehabilitation, an inability to independently adjust their psychological emotions and coping styles, a low level of psychological consistency, poor self-management ability and poor post-operative rehabilitation effects (17). The results of this study showed that the level of psychological consistency was low in the 90 patients with type A AD, suggesting that post-operative psychological consistency among type A AD patients is generally low. Therefore, it is important to analyze the relevant factors affecting the post-operative psychological consistency of

TABLE 4 | Linear regression analysis of the influence of various factors on the sense of psychological consistency of patients with type A AD.

Factor	B value	βvalue	95% CI of B value	*t*	*P*
Constant	43.333	2.365	38.632–48.035	18.321	0.001
Monthly household income (X_1)	6.667	2.897	0.909–12.424	2.301	0.024
Family function (X_2)	16.730	1.896	12.961–20.499	8.822	0.001

patients with type A AD and to take corresponding measures to improve the rehabilitation effects of patients with type A AD.

A sense of psychological consistency mainly refers to the individual's tendency to maintain self-confidence when faced with stress and pressure, which is mainly reflected in his or her understanding of stressors and pressure and perception of his or her own coping ability (18). A sense of psychological consistency includes a sense of control, a sense of understanding and a sense of meaning, in which the sense of control refers to the individual's self-confidence in dealing with problems, the sense of understanding refers to the individual's recognition of the logic of events from the internal and external environment, and the sense of meaning refers to the individual's subjective judgment and understanding of events. The three components work together to exert a positive psychological effect so that patients can correctly perceive stress and stressful events and respond positively (19). Family function is a comprehensive evaluation index of the family. On the one hand, it indicates whether the basic material needs of family members are being met, and on the other hand, it reflects the emotional communication and family cohesion between family members. It mainly refers to the overall self-perceived ability of the family to deal with pressure and suffering (20). One study noted that patients with good family function could mobilize more family resources and that family members could provide additional care, which is conducive to enhancing patients' confidence in coping with diseases and helping them change their coping styles (21). Therefore, it is preliminarily speculated that family functioning may affect patients' sense of psychological consistency. The results of multiple linear regression analysis show that family function can affect the post-operative psychological consistency of patients with type A AD. The reason is that patients with close emotional ties among family members can better express their post-operative psychological emotions. With the help of family members, patients' psychological adaptability is improved, and their acceptance of the disease is improved, which is conducive to helping patients establish confidence in rehabilitation (22). On the other hand, good family functioning is conducive to improving patients' ability to manage disease. Mutual support and the sharing of resources among family members can facilitate patients' disease management to improve patients' self-management ability, and the sharing of responsibilities can actively mobilize patients' subjective initiative and improve their sense of psychological consistency (23). Good family function can create a comfortable rehabilitation environment for patients, offer patients physiological care and psychological comfort, encourage patients' physiological and psychological rehabilitation, help patients endure pressure and stressors, reduce psychological burden, and improve patients' sense of psychological consistency (24, 25). In addition, this study also found that family monthly income can affect patients' sense of psychological consistency, which may be related to the fact that treatment and rehabilitation costs can increase patients' psychological burden on the family.

The relevant research shows that (26, 27) researchers have mainly conducted family functional intervention through cognitive behavioral therapy, family psychological education, family membership and so on. Cognitive behavioral interventions are mainly performed through health development and the distribution of manuals, health lectures, personalized communication, etc. Family psychological education is mainly based on multidisciplinary cooperation and family members' participatory care, providing rehabilitation knowledge to patients. Disease-related knowledge should be disseminated to major family caregivers, medical caregivers, nursing caregivers and family members so they can jointly formulate personalized care plans. At the same time, disease care and other related help should be provided via WeChat, telephone, the internet and other ways. Intervention with family members benefits both patients and all family members, and meetings should be held to address disease-related impacts and problems. Such impacts include the impact of the illness on family members' lives, psychological pressure and economic burden, role changes and the ability to deal with emergencies. Through different interventions, the cognition of patients and family members can be improved; the relationship, psychological state and coping ability of family members can be improved; the intimacy of family members can be increased; and family function can be improved.

CONCLUSION

In conclusion, the post-operative psychological consistency of patients with type A AD is related to family function. There are various clinical interventions for family function. It is suggested that personalized interventions be selected according to the particularities of type A AD patients to meet patients' needs for family support, mobilize family function, and improve patients' sense of psychological consistency.

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 review and approval was not required for the study on human participants in accordance with the local legislation and institutional requirements. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

XZ and JC conceived of and designed the study. XZ and YZ collected and analyzed the data. XZ drafted the manuscript. All authors have read and approved the final manuscript.

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CASE REPORT
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The Surgical Strategy for Progressive Dilatation of Aortic Root and Aortic Regurgitation After Repaired Tetralogy of Fallot: A Case Report

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It has been found that postoperative progressive dilatation of aortic root is not rare for adult patients with repaired Tetralogy of Fallot (TOF), which leads to severe aortic regurgitation or even fatal dissection. Therefore, clinically, surgical treatment for both regurgitated aortic valve and dilated root is needed based on preoperative assessments and individual treatment strategies.

Keywords: aortic root, tetralogy of Fallot, left ventricular outflow tract, cardiopulmonary bypass, cardiac surgery

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INTRODUCTION

Clinically, how to manage and prevent structural abnormalities for both aortic valve and root after repaired TOF (rTOF) has been the predominant point in the surgical field, although, it has been reported that compared with the incidence of postoperative aortic aneurysm, which is the dilatation of aortic root, is relatively lower (12 vs. 9%) (1). Currently, fewer case reports of surgical treatment focusing on how to handle postoperative dilatation of aortic root and aortic regurgitation (AR) after smoothly and reasonably has been shown (2, 3). For our case report, the David I plus repaired aortic valve procedure, combined with simultaneous reconstruction of the left ventricular outflow tract (LVOT), were selected as the surgical option by individual preoperative assessments, and the postoperative outcome of this patient is satisfactory. Hence, it is valuable to share our surgical experience as follows.

CASE PRESENTATION

A 39-year-old male patient was admitted, complaining of persistent hemoptysis for 3 weeks. The patient was a typical TOF, not TOF of SD I type. Previously, he underwent radical surgery for tetralogy of Fallot, including relief of right ventricular outflow tract obstruction and repair of ventricular septal defect at 21. A diastolic murmur was found in the second left intercostal space of the sternum and other physical examinations were negative. Based on findings observed from cardiac echocardiography, it was diagnosed as dilatation for both sinotubular junction and ascending aorta (annulus, 3.17 cm; sinus, max. 5.82 cm; proximal ascending aorta, 4.14 cm) and aortic valve insufficiency complicated with severe AR (**Figure 1A**). The preoperative thoracic CT scan showed the diameter of the aortic root was 55.19 mm × 67.09 mm (**Figure 2**). Meanwhile, local obstruction of LVOT was also detected (**Figure 1B**), the gradients of LVOT obstruction was 23 mmHg. After clinical assessments, without any contradictions, this patient received the procedure including both David I plus repaired aortic valve procedure, combined with simultaneous reconstruction of LVOT.

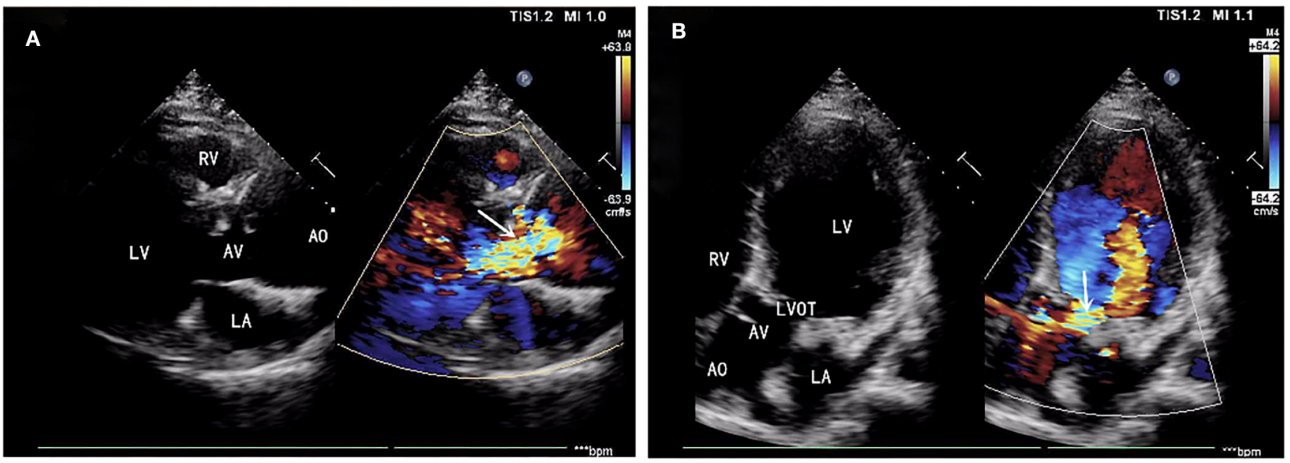

FIGURE 1 | (A) Aortic valve insufficiency complicated with severe aortic regurgitation (AR). **(B)** Local obstruction of left ventricular outflow tract (LVOT) (gradients of LVOT 23 mmHg). AR, aortic regurgitation; LVOT, left ventricular outflow tract.

Firstly, the separation of both femoral artery and vein was completed through the incision of the right groin. The median sternal incision was then taken and the separation for both superior and inferior vena cava was completed. Sequentially, the cardiopulmonary bypass (CPB) was established after stable catheterization into femoral artery and vein, as well as superior vena cava. The ascending aorta was clamped and Histidine-Tryptophan-Ketoglutarate solution (HTK) Cardioplegia was initiated at 34°C. Transection of the aorta at 1 cm above the sinotubular junction. Visually, dilatation of the aortic annulus and enlargement of the sinus were found. The aortic valve was not thickened, and leaflets were well-preserved. However, due to the dilatation of the aortic root, leaflets were mismatched with the aortic annulus, which will lead to misalignment of the aortic valves. After evaluating the function of the aortic valve, the thickened muscle bundles were seen in the left ventricular outflow tract *via* the aortic valve, and the thickened muscle bundles were removed with a fish abdominal blade. Then, the aortic root was separated for the measurement of the distance between the commissure of three leaflets and the annulus. Also, the distance between the midpoint and root of three leaflets was calculated at the same time. The aortic root, 3 mm above the annulus, was cut (**Figure 3A**) and a 32-mm artificial graft was selected. The height for commissure of leaflets was adjusted reasonably and each cusp of leaflets was fixed within the artificial graft by horizontal mattress suture during proximal anastomosis (**Figure 3B**). Anastomosis for both left and right coronary arteries to graft was completed by button technique. Lastly, distal anastomosis between the aorta and graft was performed. The ascending aorta was declamped after enough rewarming, the physiological heart rhythm was then activated automatically and smoothly. Then, no significant AR was found through intraoperative esophageal ultrasound. CPB was weaned when hemodynamics and vital signs were stable. Both regulatory chest closure and drainages for mediastinum of left and right chest cavities were performed without any active bleeding. The patient was transferred to the ICU ward for further medical supplements after surgery. The postoperative extubation was completed within the first 12 h in the ICU ward. It was detected that the size and shape of the graft were satisfactory and the function of the aortic valve was normal with mild regurgitation. Later, III° atrioventricular block was found by dynamic electrocardiogram, therefore, based on the consultant from a senior cardiologist and with the aim for improvement of rhythm, implantation of a permanent pacemaker was performed. Finally, the patient discharged after systematic postoperative clinical assessments were stable and qualified. Postoperative pathology showed atrophy and degeneration of smooth muscle in media of the aortic wall, focal vitreous degeneration, myxoid degeneration, and calcium salt deposition (**Figure 4**). The follow-up echocardiogram 2 months after the operation showed significant improvement in local obstruction of LVOT (Gradients of postoperative LVOT 7 mmHg), with trivial aortic regurgitation (**Figure 5**). During the follow-up of 1 year after surgery, the surgical effect is still positive and confirmed without any significant worse progression.

DISCUSSION

In a prospective trial, larger LV stroke volume has been found as an independent risk factor for delayed dilatation of aortic root due to increased shear stress (4). Based on the findings from 4D flow MRI, it has been suggested that among the patients after rTOF, elevated stiffness for both ascending and descending aorta is more significant and the wall shear stress (WSS) increases following physical growth. Although the diameter of the aorta remains in the normal range for a short time after the first repair of TOF, late dilatation of the aortic root and damage to the aortic wall are still inevitable (5). Moreover, up to the median of 7 years since the first repair, aortic stiffness is related to the incidence and severity of aortic root dilatation

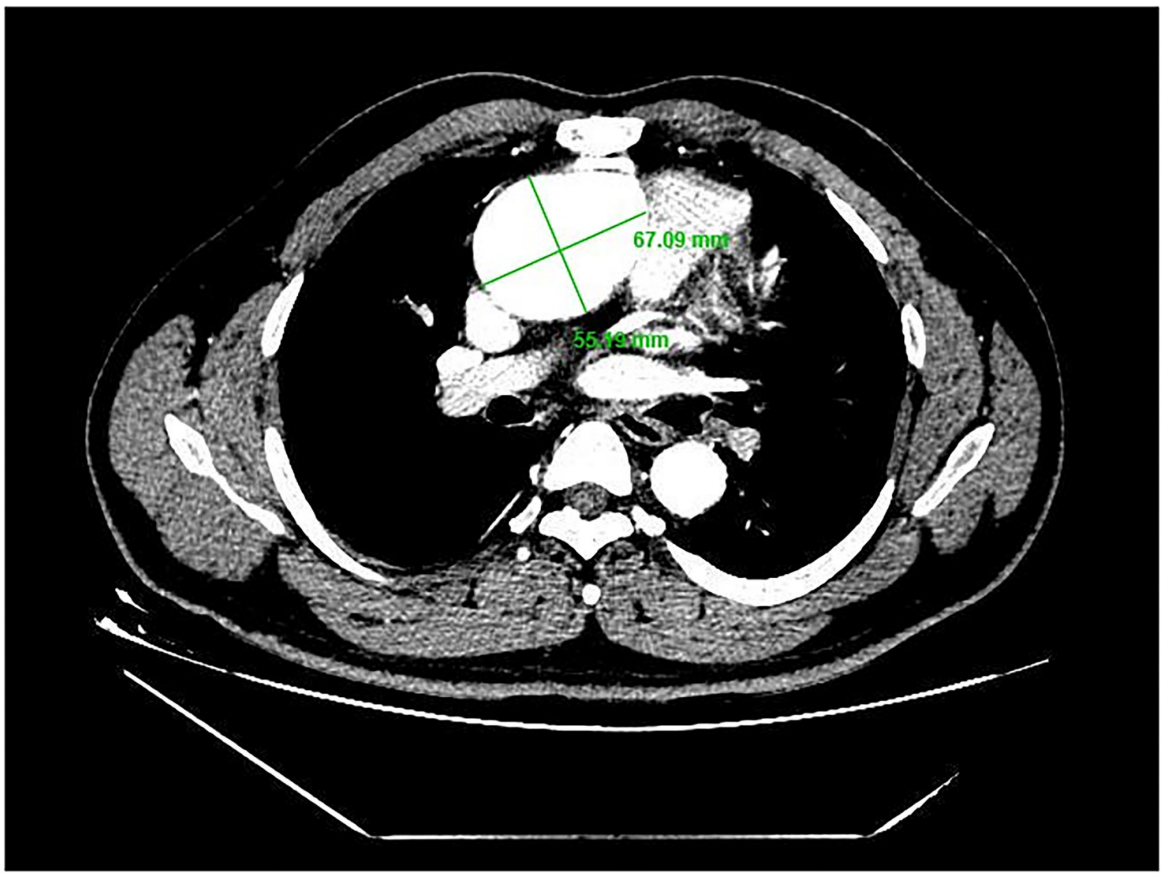

FIGURE 2 | Diameter of aortic root 55.19 mm × 67.09 mm.

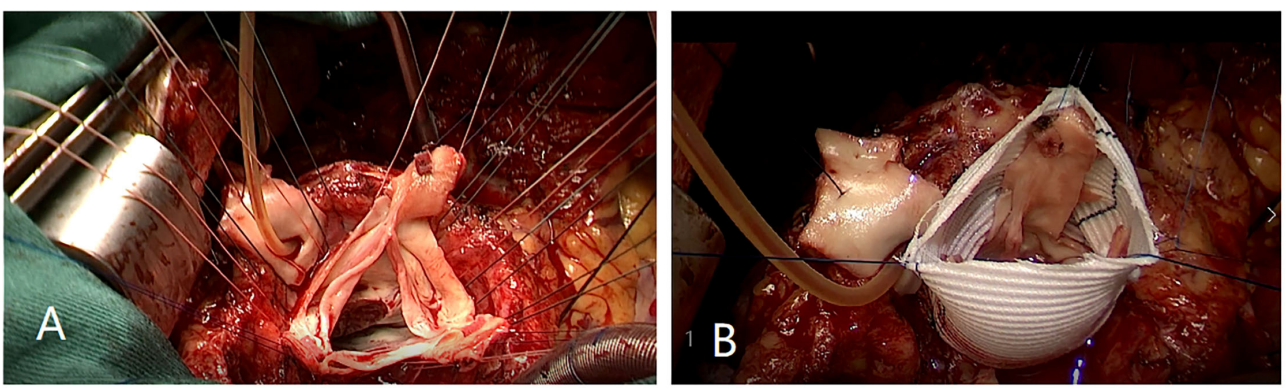

FIGURE 3 | (A) Aortic root was cut 3 mm above annulus. **(B)** Each cusp of leaflets was fixed within the artificial graft by horizontal mattress suture during proximal anastomosis.

(6). Besides, François and colleagues, in their study to explore the histologic alterations of aortic root for infants after rTOF, have found in all selected samples of aorta, fibrosis, increased mucoid accumulation, and elastin fragmentation account for 45, 15, and 5%, respectively, and within 2 years after repairing, the regression of aortic diameters is significant (7). Chowdhury et al. (8) described that both substantial lamellar loss and intrinsic aortopathy were considered critical factors to the development of aortic abnormalities including degeneration, regression, and dilatation, and it was supposed that histologic alterations were correlated with the remodeling of ascending aorta. In this case, it had been 18 years for the patient in our report since initial TOF

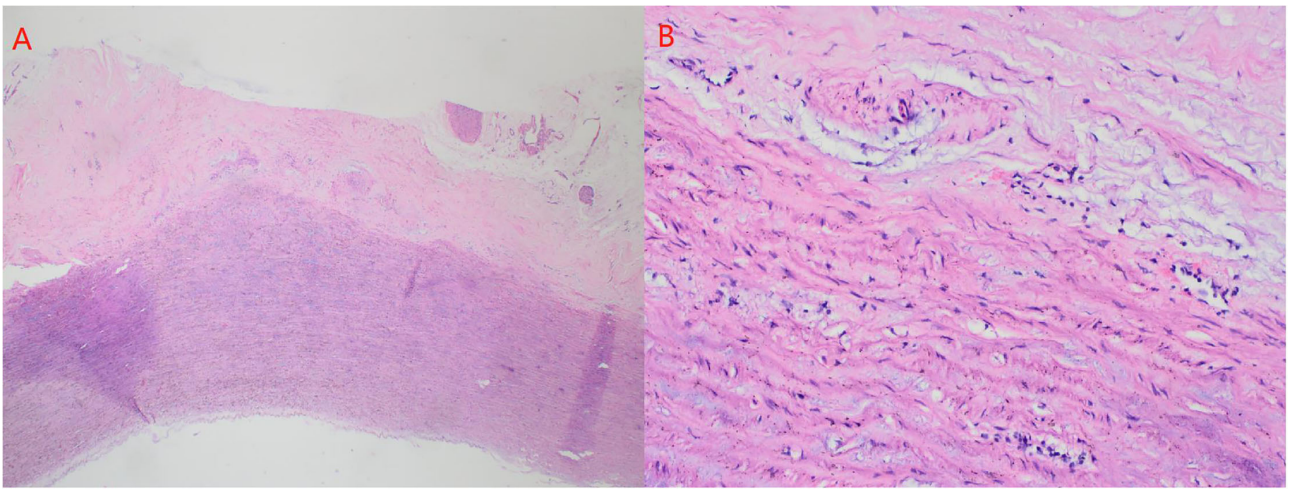

FIGURE 4 | (A) atrophy and degeneration of smooth muscle in media of aortic wall (A × 50, hematoxylin, and eosin stain). **(B)** focal vitreous degeneration, myxoid degeneration (B × 200, hematoxylin, and eosin stain).

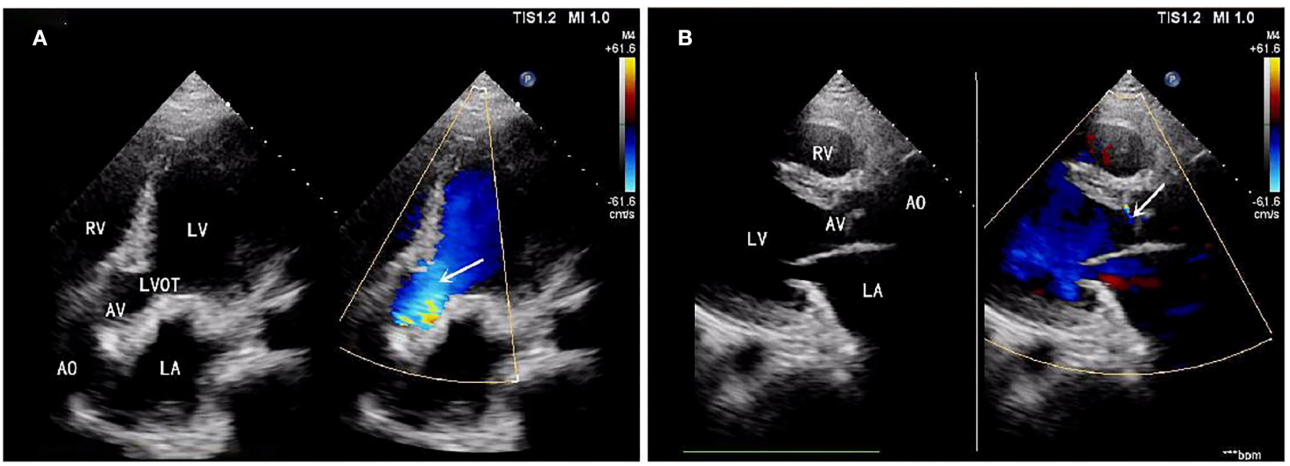

FIGURE 5 | (A) Left ventricular outflow tract obstruction disappeared (Gradients of LVOT 7 mmHg). **(B)** Trivial aortic regurgitation.

repairing, therefore, the structural abnormalities of the aortic root were progressive under the persistent negative influence of WSS. Meanwhile, the dysfunction and hypertrophy of LV were also presented due to elevated afterload and abnormal LV flow tract. Possibly, stiffness, as one explained the mechanism of aortic dilatation, reduces the flexible luminal expansion, and leads to elevated WSS, then, finally, induces turbulent flow within the aorta. Postoperative pathological results also showed atrophy and degeneration of aortic medial smooth muscle, focal vitreous degeneration, myxoid degeneration, and calcium salt deposition (**Figure 4**). In terms of organization and structure, this also confirms our assumption about the mechanism of progressive dilatation of aortic root after the operation of tetralogy of Fallot in this patient. We believe that the postoperative progressive dilatation of aortic root after rTOF is the leading cause for postoperative AR mainly associated with altered hemodynamics and inherent defects of the aortic wall. During the follow-up period, aortic dilatation has been found in most adult patients with rTOF, interestingly, however, it is concepted that as a part of normal physical process, not all aortic dilatation cause AR inevitably (9). This may be due to the dilatation of the aortic root did not involve the aortic annulus. It was worth mentioning that this patient performed CTA but not MRI preoperative. However, in a prospective cardiovascular MRI study, it has been recommended that as a comprehensive and accurate screening tool, with the aim of preventing TOF-associated aortic complications, cardiovascular MRI after rTOF is more necessarily needed to assess the diameter of the aortic root and detect residual of outflow tract, as well as structural data of right and left ventriculars (10).

Commonly, surgical intervention is the primary option when the diagnosis of aortic complications after rTOF

is confirmed. Whereas the risks for both postoperative complications and early mortality are significantly higher, therefore, preoperative assessment should be emphasized (11). Currently, the recommended threshold of ascending aorta for surgical intervention is set as 55 mm (12). Bentall procedure is now the regular surgical skill for medical intervention of aortic complications with the advantages of improvement of LV function and decreased LV mass based on findings through long-term follow-up (13). Valve–sparing aortic replacement (VSRR) is an effective and safe surgical technique for pediatric patients with an aneurysms. Compared with the Bentall procedure and bioprosthetic valve conduit (BVC) replacement, the outcome after VSRR shows similar excellent survival and freedom from aortic re-intervention rates up to 10 years (14). Nevertheless, it is emphasized that the aortic valve can be spared instead of replaced mechanical or biological prosthesis, hence, both anticoagulation and degeneration of replaced prosthesis are avoided reasonably (15, 16). David I-V procedures are updated from the original VSRR. For this younger patient, David I plus repaired aortic valve procedure, combined with simultaneous reconstruction of LVOT, was selected after preoperative assessment and individual considerations, which could be an ideal and radical solution for aortic diseases with lower mortality and prevent adverse events after surgery, especially for anticoagulation-associated bleeding.

CONCLUSION

Long-term follow-up of aortic root and valve is necessary for patients with rTOF. Any aortic complications, such as dilatation of aorta root or ascending aorta, and AR can be detected by imaging MRI. As an alternative skill for both Bentall and BVC, David's procedure is a flexible and critical component for surgical intervention to improve dilatation of aortic root and severe AR after rTOF, which maintains stable hemodynamics within the aorta and morphological characteristics of the aorta and without anticoagulation-associated serious adverse events.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/**Supplementary Materials**, 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

SZ and HL: collected relevant data and drafted the manuscript. XW and SH: taking pictures. CZ and HL: revised the manuscript. CZ and SZ: supervised the audit process. All authors read and approved the final manuscript.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fcvm.2022.840946/full#supplementary-material

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