

# Exploration to the Application of Hybrid Technology in Special Types of Congenital Heart Diseases in Children

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## Research article

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# Abstract

## Background

Hybrid technology has become a hot topic in cardiovascular surgery, has been widely used in the minimally invasive closure of simple coronary heart diseases (CHDs). For some children with special CHDs, it is still impossible to avoid the huge trauma caused by cardiopulmonary bypass. This study aimed to investigate the feasibility, safety and efficacy of hybrid technology in the treatment of several specific CHDs.

## Methods

A total of 29 children with specific CHDs hospitalised in the Cardiac Surgery Department of Dalian Children's Hospital from July 2014 to June 2019 were enrolled. There were 2 cases of right coronary artery-right ventricular fistula, 17 cases of neonatal critical pulmonary stenosis (CPS), 9 cases of neonatal pulmonary atresia-intact ventricular septum (PA/IVS), and 1 case of giant aortopulmonary window (APW). All of them underwent surgical treatment with hybrid technology guided by transoesophageal echocardiography (TEE). The TEE enabled immediate evaluation of the surgical curative effect. Further chest X-ray, electrocardiogram (ECG) and echocardiogram were performed in the outpatient department after discharge.

## Results

The surgical treatment with hybrid technology was smooth except that 1 CPS patient was converted to open-heart surgery with cardiopulmonary bypass (CPB) due to a torn right ventricular outflow tract after balloon dilatation. No complication, such as wound or intracardiac infection, arrhythmia or pericardial effusion occurred. No children have been lost to follow-up and the investigation results and prognosis remain satisfactory.

## Conclusions

the use of hybrid technology is a safe and effective alternative therapy for specific paediatric CHD cases. It has significant advantages in alleviating trauma and reducing medical costs and, therefore, has good prospects for broad application in the future.

## Introduction

Hybrid technology is a new composite technology integrating surgical intervention, imaging and other disciplines<sup>[1]</sup>. The application of hybrid technology to cardiovascular surgery is an organic combination

of interventional technology and surgery, giving full play to the advantages of both<sup>[2]</sup>. It retains the advantages of safe and minimally invasive interventional technology, but is not limited by the patient's weight and blood vessels, and limits radiation exposure<sup>[3]</sup>. Hybrid technology has been widely used in the minimally invasive closure of simple CHDs, such as ventricular septal defect (VSD), atrial septal defect (ASD), and patent ductus arteriosus (PDA) <sup>[4]</sup>. Over the past decade, interventionalists have accumulated a significant degree of clinical experience. With the rapid development of cardiac diagnosis and neonatal cardiac surgery, the treatment of CHD tends to be at a younger age and can be more complicated<sup>[5]</sup>. Traditional CPB cannot meet the urgent needs of minimally invasive treatment for some children with special CHDs. Hybrid technology, as an alternative, has become a hot topic in the field of cardiovascular surgery<sup>[6]</sup>. The Cardiac Surgery Department of Dalian Children's Hospital explored the use of Hybrid technology in the treatment of some specific types of CHD as a substitute for open-heart surgery under CPB from July 2014 to June 2019 and obtained good results. The experience is reported here.

## Patients And Methods

Twenty-nine children with specific CHDs admitted to the Cardiac Surgery Department of Dalian Children's Hospital from July 2014 to June 2019 were enrolled for this study, including 2 cases of right coronary artery-right ventricular fistula, 17 cases of neonatal critical pulmonary stenosis (CPS), 9 cases of neonatal pulmonary atresia-intact ventricular septum (PA/IVS), and 1 case of giant aortopulmonary window (APW). Informed consent was signed by the guardian before operation.

## Coronary Artery-right Ventricular Fistula

Two children had a right coronary artery-right ventricular fistula. The first case was a female of 8 months weighing 9.2 kg, and the second case was a male of 10 months weighing 9.7 kg. Both infants were diagnosed with cardiac ultrasound and had grade II cardiac function.

Surgical method: We placed the children in the supine position, performed tracheal intubation and routine anaesthesia, and inserted the TEE probe to detect the location and blood flow of the right ventricular fistula (Fig. 1A. Figure 2A.). A small 3 cm incision in the median lower sternum was made, and the pericardium was opened and suspended. The enlarged right coronary artery could be seen, (Fig. 3). A VSD occluder with a waist size appropriate for the fistula size was selected (a 5 mm symmetric VSD occluder was used in both children). After the occluder was in place, the TEE was used to show that its shape was satisfactory, without residual bypass (Fig. 2B. Figure 2B.). ECG monitoring showed there was no arrhythmia or elevated T-wave and the push-pull test indicated that the umbrella was firm and stable; the occluder was then released. A drainage tube was secured in the pericardial cavity, and we performed routine sternal closure.

## Neonatal Cps And Neonatal PA/IVS

Neonatal CPS and neonatal PA/IVS are discussed together despite the different haemodynamics and pathophysiology as they share a similar surgical procedure. Among the 17 cases of neonatal CPS and 9 cases of neonatal PA/IVS, the mean age of the infants was  $10.5 \pm 2.8$  days, the operation body mass was  $3.7 \pm 0.9$  kg, the inner diameter of the pulmonary valve ring was  $7.3 \pm 1.4$  mm, and the tricuspid Z value was  $-1.9 \pm 0.6$ ; the right ventricular development of all children was mild-moderate according to the auxodrome put forward by Bull<sup>[7]</sup>, excluding the PA/IVS with severe tricuspid Ebstein's malformation or severe right ventricle dependent coronary circulation. The ductus arteriosus remained open in all children as a result of intravenous administration of vitamin PGE1.

**Surgical method:** We placed the child in the supine position, performed tracheal intubation and administered routine general anaesthesia. The inner diameter of the pulmonary valve ring, the size of the tricuspid valve ring and the development of the right ventricle were verified by TEE. A mid-sternal incision was made and the pericardium suspended, exposing the outflow tract of the right ventricle and pulmonary artery. We then performed a purse-string suture in the outflow tract of right ventricle 2.0 cm from the pulmonary valve, punctured the outflow tract of the right ventricle with an 18G puncture needle, and connected the manometer tube to measure the transvalvular gradient between the right ventricle and pulmonary valve, and recorded it. We then placed a guidewire in a 7F sheathing tube through the pulmonary valve guided by TEE and withdrew the guidewire. We chose a balloon 1.2 times the inner diameter of the pulmonary valve ring, and dilated it 3 times, for 3 s each time (Fig. 4A, B, C, D). Note: (1) the upper 2/3 of the balloon should be above the pulmonary valve ring to avoid damage to the outflow tract of the right ventricle; (2) the sheathing tube should be fixed during balloon dilation, to avoid balloon displacement and damage to the right ventricular outflow tract. After balloon dilation, the right ventricular pressure should be measured again with the right heart catheter. Myocardial thickening or spasm of different degrees narrowed the right ventricular outflow tract after the surgery, resulting in a low oxygen partial pressure. Therefore, we ligated the ductus arteriosus in 7 children with large ductus arteriosus after BT bypass and maintained the PGE1 infusion to delay ductus arteriosus closure as long as possible.

## Giant APW

The cases of giant APW were male children aged 4 months, with a weight of 6.6 Kg, who were hospitalised due to recurrent respiratory tract infections, difficulty in feeding, failure to increase weight, excessive sweating and progressive shortness of breath. The colour Doppler ultrasound showed that in the artery short shaft section, there was no detected echo signal between the two main arteries at the short axial section of the aorta. The missing length was about 11 mm, and this case was diagnosed as type II APW according to Richardson's classification<sup>[8]</sup>, which was further confirmed by computed tomography angiography.

**Surgical method:** The child was placed in a supine position to undergo endotracheal intubation. TEE showed an enlarged left atrium and ventricle, and low-moderate reflux in the mitral valve, with a reflux bundle at the junction of the valve leaflet, which was caused by the left ventricular enlargement resulting

from the APW. By gently pulling the probe outward into the horizontal section of the aortic arch, adjusting the angle to 10 °–20 °, and gently lifting the probe again, the APW could be displayed. It was 11 mm long after measurement (Fig. 5. A), which was consistent with the results of the preoperative diagnosis. This was the first attempt to apply hybrid technology to occlude an APW. To minimise the surgical risk, a conventional midline incision was made to free the aortic arch, main pulmonary artery septum and left and right pulmonary arteries. The extracorporeal circulation machine was kept ready. Due to the large APW defect, the direct use of a large-size VSD occluder could block the blood flow of aorta and right pulmonary artery. Therefore, the surgeons partially ligated the APW to create an inside diameter of 6 mm under the guidance of TEE and then used a 6 mm symmetrical equilateral VSD occluder in the ligated APW. Closure was achieved by performing a purse-string suture at the root of the main pulmonary artery, placing the puncture needle and then withdrawing the needle core. The cannula was advanced through the APW under the guidance of TEE, then withdrawn. The transfer sheathing was advanced through the APW along the guidewire, and the guidewire was withdrawn. TEE showed a 'dual-track' sign of the sheathing tube (Fig. 5. B). When mounting the occluder, we first released the aorta-side umbrella, pulled it back close to the defect edge, continued to withdraw the sheathing tube to release the umbrella plate on the pulmonary artery side, made the left and right umbrella plates clamp the defect edges, and the waist clamped the partially ligated APW(Fig. 5.C). After occlusion, TEE was used to comprehensively evaluate the release of the umbrella and any residual bypass. Color doppler showed normal flow rates in the left and right pulmonary arteries and the aortic arch, confirming that the occluder had created no significant effect on the blood flow of the aorta and right pulmonary artery(Fig. 5. D). After making sure the occluder was firm by pushing and pulling the steel wire, we withdrew the wire and sheathing tube, tightened and tied off the purse-string suture. We gave protamine (1:1) and heparin. After achieving haemostasis, we closed part of the pericardium, placed the drainage tube and performed sternal closure in layers.

## Results

The two children with coronary right ventricular fistula were treated with oral aspirin only for anticoagulation, cardiac diuretics or sedatives were not used. After the ECG, chest X-ray and colour doppler ultrasound examinations at follow-up, no arrhythmia, myocardial ischaemia, residual bypass, occluder displacement or thrombosis occurred, and cardiac function was significantly improved.

Among the 17 neonates with CPS, one case was converted due to laceration of the right ventricular outflow tract. This resulted from displacement during balloon dilation in one case due to inexperience in the initial stage. The other 16 patients underwent hybrid technology procedures successfully. Immediately after balloon dilation, the pressure difference across the pulmonary valve decreased from ( $108.3 \pm 16.6$ ) mmHg to ( $33.7 \pm 11.4$ ) mmHg ( $P < 0.001$ ), the right ventricular systolic pressure decreased from ( $119.7 \pm 16.3$ ) mmHg to ( $45.2 \pm 10.5$ ) mmHg, and the ratio of right ventricular pressure to aortic pressure decreased significantly ( $1.6 \pm 0.3$  before surgery,  $0.8 \pm 0.2$  after surgery,  $P < 0.001$ ). TEE showed that the pulmonary valve opening after balloon dilation was significantly improved and tricuspid regurgitation was significantly reduced. The duration of postoperative mechanical ventilation was ( $4.5 \pm 1.6$ ) days, and the mean hospital stay was ( $11.3 \pm 4.1$ ) days. At discharge, the mean oxygen saturation of 16 children

rose to 87.5%. The 16 children were followed up for  $32.5 \pm 6.5$  months, and ultrasonography showed mild pulmonary valve regurgitation in 3 cases and moderate regurgitation in 1 case. 16 patients underwent biventricular radical resection and one patient underwent a semi-ventricular correction in stage II, while 3 patients are awaiting stage II surgery.

The procedures for the 9 patients with PA/IVS were successful, with a mean duration of 112 minutes (range 95–147) minutes. The balloon size was 6–10 mm (mean on average). One child died of severe pulmonary infection, and the remaining 8 children were discharged from hospital successfully. The mean duration of mechanical ventilation of the 8 surviving children was 31 hours (range 22–47)), the mean duration of hospitalisation was 17 days (12–23 days), and the mean oxygen saturation of the 8 children rose to 87.5%, the mean right ventricular systolic blood pressure dropped to 49 mmHg, and the mean transvalvular pressure of the pulmonary valve was 28 mmHg. One patient developed transient supraventricular tachycardia after surgery and recovered without treatment. Five children were given a small blood transfusion due to intraoperative blood loss, 3 children had a minor degree of pulmonary valve regurgitation, 1 child had a small amount of pulmonary valve regurgitation, and no one suffered from cardiac perforation, aortic aneurysm or low cardiac output syndrome. The 8 children were followed up for a mean of 28.5 months (range 7–56 months). Five children have received radical surgery, and three children are waiting for radical surgery.

The occlusion process in the children with giant APW was smooth, and intraoperative TEE showed that the APW bypass disappeared, the push-pull test confirmed that the occluder was firm and stable, the left atrium and ventricle were significantly retracted after the surgery, and the mitral valve regurgitation was significantly reduced. The left third intercostal space murmur disappeared at auscultation. Echocardiography showed that the position of the occluder was satisfactory and stable after 3 months of follow-up. Echocardiography showed that the volume of the left atrium, left ventricle and pulmonary artery pressure gradually returned to normal after 6-months of follow-up.

## Discussion

Congenital heart disease (CHD) is a common congenital malformation, which is mainly caused by abnormal development of large blood vessels of the heart in the fetal period. According to statistics, its incidence is about 0.7–0.8%<sup>[9]</sup>. Open-heart surgery under traditional cardiopulmonary bypass (CPB) and percutaneous intervention are the two main strategies in the treatment of CHD<sup>[10–13]</sup>. However, CPB can lead to a series of complications, and the indications of interventional therapy are relatively strict. The application of both is greatly limited<sup>[14–16]</sup>.

Angelini proposed the concept of hybrid intervention for the first time and successfully used it in the treatment of coronary heart disease in 1996<sup>[17]</sup>. As a part of minimally invasive cardiac surgery, Hybrid intervention is the composite product integrating intervention and surgery in an organic manner, which takes advantages of both to tailor the best therapy for patients<sup>[18]</sup>. It has been widely recognised and promoted in the various branches of cardiovascular surgery owing to its significant advantages. At

present, the treatment of simple CHD, such as ASD, VSD, PDA, PS, for example, has been gradually popularised and significant clinical experience has now been accumulated. Nevertheless, the treatment of CHD tends to be in younger-aged patients and is further complicated by the rapid development of fetal cardiac diagnosis and neonatal medicine. For some specific congenital heart diseases in children<sup>[19]</sup>, conventional cardiac operation under CPB become more and more difficult to meet the urgent requirements of patients for minimally invasive treatment. and it has become a hotly debated question in the field of cardiovascular surgery, whether hybrid technology can be used as an alternative therapy<sup>[20]</sup>.

Since 2011, the Cardiac Surgery Department of Dalian Children's Hospital has treated thousands of patients with ASD, VSD, PDA, PS and other simple CHDs by hybrid-assisted minimally invasive procedures with a small transthoracic incision, guided by TEE. We have accumulated rich clinical experience. The department, therefore, made an attempt to explore the use of hybrid technology in the treatment of children with some specific CHDs, including 2 cases of right coronary artery-right ventricular fistula, 17 cases of neonatal CPS, 9 cases of neonatal PA/IVS and 1 case of APW. The cases above show that hybrid technology has the following advantages: (1) it expands the therapeutic indications, and effectively bypasses the restrictions of the traditional methods imposed by age, weight, vascular conditions and cardiac anatomical structure; (2) there is no need for CPB or it minimises CPB time, reduces the damage to important organs caused by the systemic inflammatory response, and avoids or reduces the exposure of children to X-rays; (3) it reduces or eliminates blood transfusions; (4) some hybrid treatments are performed under direct vision, and this can avoid the complications associated with indirect vision; (5) intraoperative hybrid treatment is often performed by cardiothoracic surgeons in the operating room. In case of difficulties or accidents, it is a more convenient way to provide emergency treatment, and the patient can be quickly transferred to traditional CPB surgery, to guarantee higher safety.

TEE-guided hybrid surgeries create a greater need for ultrasound guidance. Experienced sonographers will need to work closely with surgeons, communicating about the real-time ultrasound images in a fast, clear and accurate manner. They need to inform surgeons of the positions of the guidewire and catheter, to avoid accidentally damaging valves or other important structures during surgery. Surgeons should also acquire the knowledge of ultrasonic imaging and master the skill of performing open-heart surgery under CPB with minimal delay.

TEE can provide particularly good guidance, but it is still invasive. The sonographers should clearly understand the indications and contraindications of TEE, and comprehensively evaluate patients beforehand. Patients with space-occupying lesions in the oesophagus, oesophageal diverticula, congenital malformations of the oesophagus, congenital or acquired upper gastrointestinal diseases, such as active upper gastrointestinal haemorrhage, oesophageal lacerations, perforated hiatal hernia, a recent history of oesophageal surgery, oesophageal varices or pharyngeal abscess should be excluded from TEE-guided surgery. Even for CHD patients without these contraindications, there is a risk of occasional serious complications during the operation, so necessary preventive and rescue measures

should be taken in advance. Intraoperative guidance should be gentle and excessive force should be avoided.

It is important to expedite the exploration of how to expand the use of minimally invasive treatment, to reduce operative trauma to patients, and improve the safety of surgery. The application of hybrid technology has given people inspiration and provided new opportunities. With the continuous progress of medical imaging technology and medical devices, the boundaries between the fields of internal medicine and surgery are gradually disappearing, and multi-disciplinary collaboration and composite technology are more extensive. The division of physicians and surgeons will become more blurred, and the advantages of compound medical talents will be highlighted. The author believes that the progress of surgical technology does not require drastic changes. Small benefits, such as avoiding CPB, shortening operation time, shortening anaesthesia time and avoiding or reducing surgical incisions, all represent significant progress. For example, a Chinese scholar once put forward the idea of occluding the defect in ventricular septal muscle under CPB with an arrested heart. Such preliminary exploration was replaced by the possibility of puncturing through the right ventricular wall without the need for CPB. Now, the implantation of an occluder needs less time than a manual suture and shows good efficacy in the treatment of Swiss cheese-like defects. This approach can greatly shorten the CPB and operation time, and now provides a basis for achieving the occlusion by puncturing through the right ventricular wall without the need for CPB in the near future.

Creative thinking is the driving force behind medical progress, and we hope to inspire and encourage medical workers through the use of hybrid technology in the treatment of some specific types of congenital heart disease. With a better understanding of the clinical application of hybrid technology, it will be more widely used in various fields of clinical medicine, and patients will begin to benefit from increasingly individualised treatment. The author proposes three areas for investigation of the use of hybrid technology in CHD: (1) how to combine surgery more closely with ultrasound and DSA; (2) how to further design and improve surgical instruments to suit hybrid techniques; (3) how to expand its use in complicated CHD.

## Conclusion

In conclusion, as a result of multidisciplinary integration, hybrid technology has the prospect of broader use in the treatment of CHD, and it will be one of the directions of rapid development in cardiovascular surgery in the future. More clinical studies and accumulated experience are still needed.

## Abbreviations

CHDs: Congenital heart diseases; CPS: Critical pulmonary stenosis; PA/IVS: Pulmonary atresia-intact ventricular septum; APW: Aortopulmonary window; TEE: Transoesophageal echocardiography; ECG: Electrocardiogram; CPB: Cardiopulmonary bypass; VSD: Ventricular septal defect; ASD: Atrial septal defect; PDA: Patent ductus arteriosus

# Declarations

## Acknowledgements

Not applicable.

## Authors' contributions

Drs. Yuhang Liu and Ning Wang contributed to data collecte and editing the manuscript. Drs. Yuhang Liu and Ping Wen participated intreating the patients preoperatively and postoperatively. Dr Ning Wang performed the echocardiography (including TTE and TEE), Dr Ping Wen contributed to the revision of the manuscript. All authors have approved the final draft of the manuscript.

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## Availability of data and materials

The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request

## Ethics approval and consent to participate

The study protocol was approved by Dalian Children's Hospital.

## Consent for publication

All authors of this paper have read and approved the final version submitted.

## Competing interests

The authors declare no conflict of interest.

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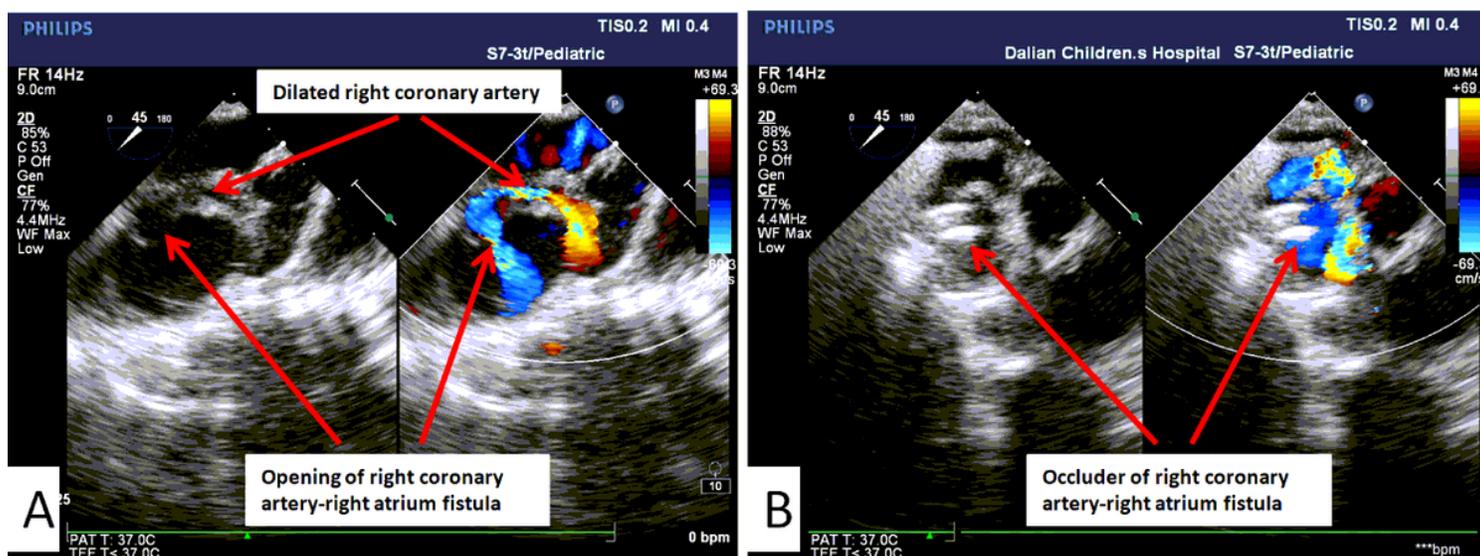
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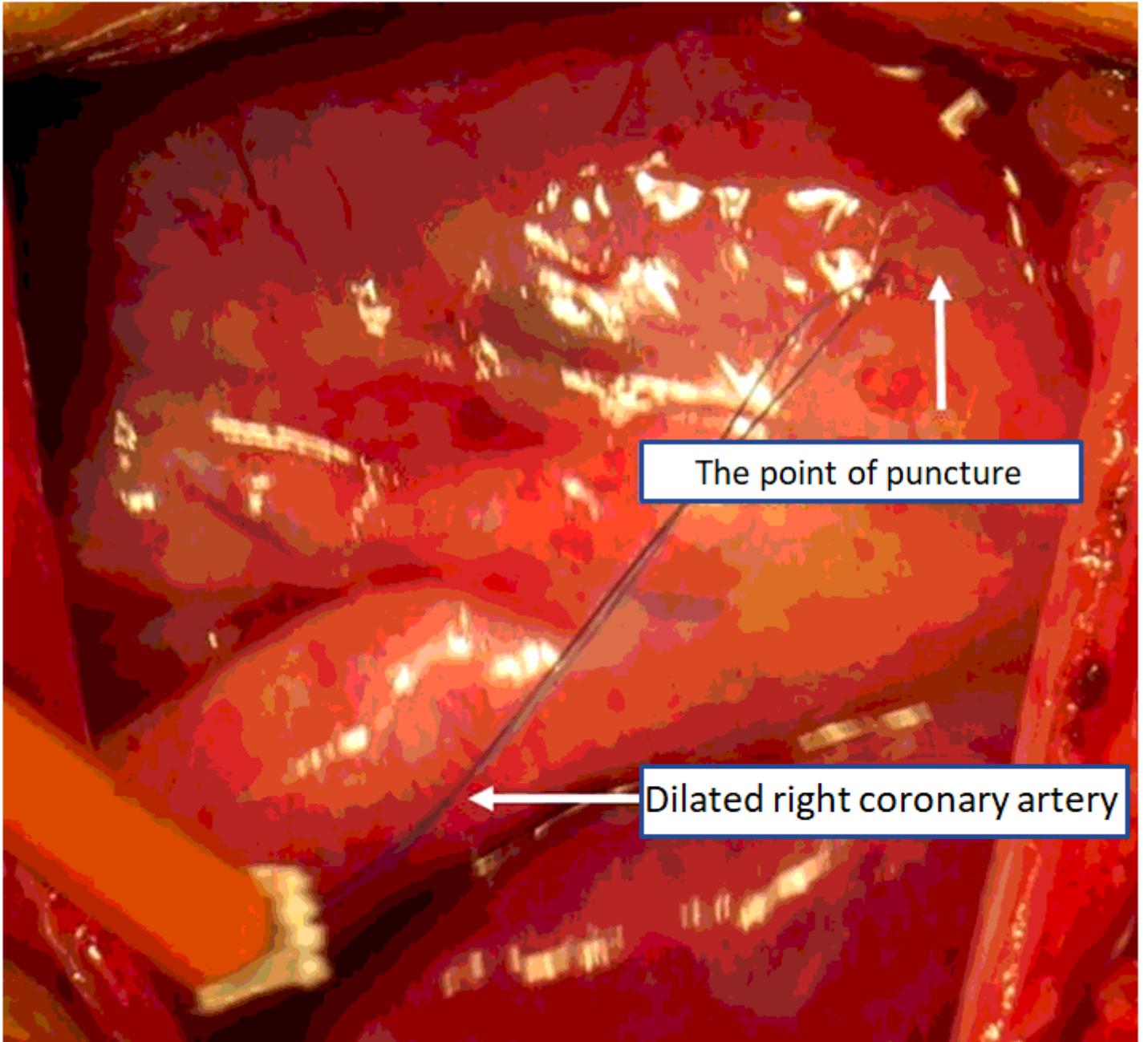
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## Figures



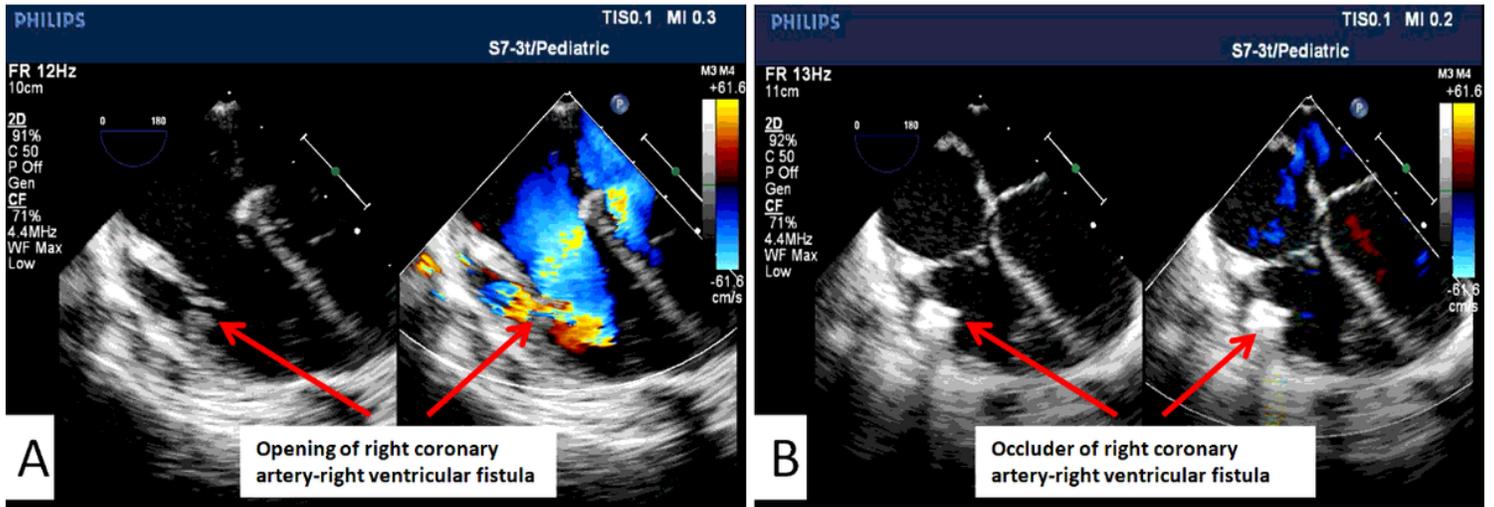
**Figure 1**

A. the right coronary artery was obviously thickened with inner diameter of 7mm, The right coronary artery backward ran above the top of right atrium and opened into the right atrium near the superior vena cava , the inner diameter is 5mm, Shunt pressure difference is 25mmHg. Left coronary artery inner diameter is 2.2mm. B.The intraoperative ultrasound of right coronary artery-right ventricular fistula after transesophageal echocardiography closure: the umbrella opened successfully, securely closed, right coronary artery - right ventricular fistula without residual shunt.



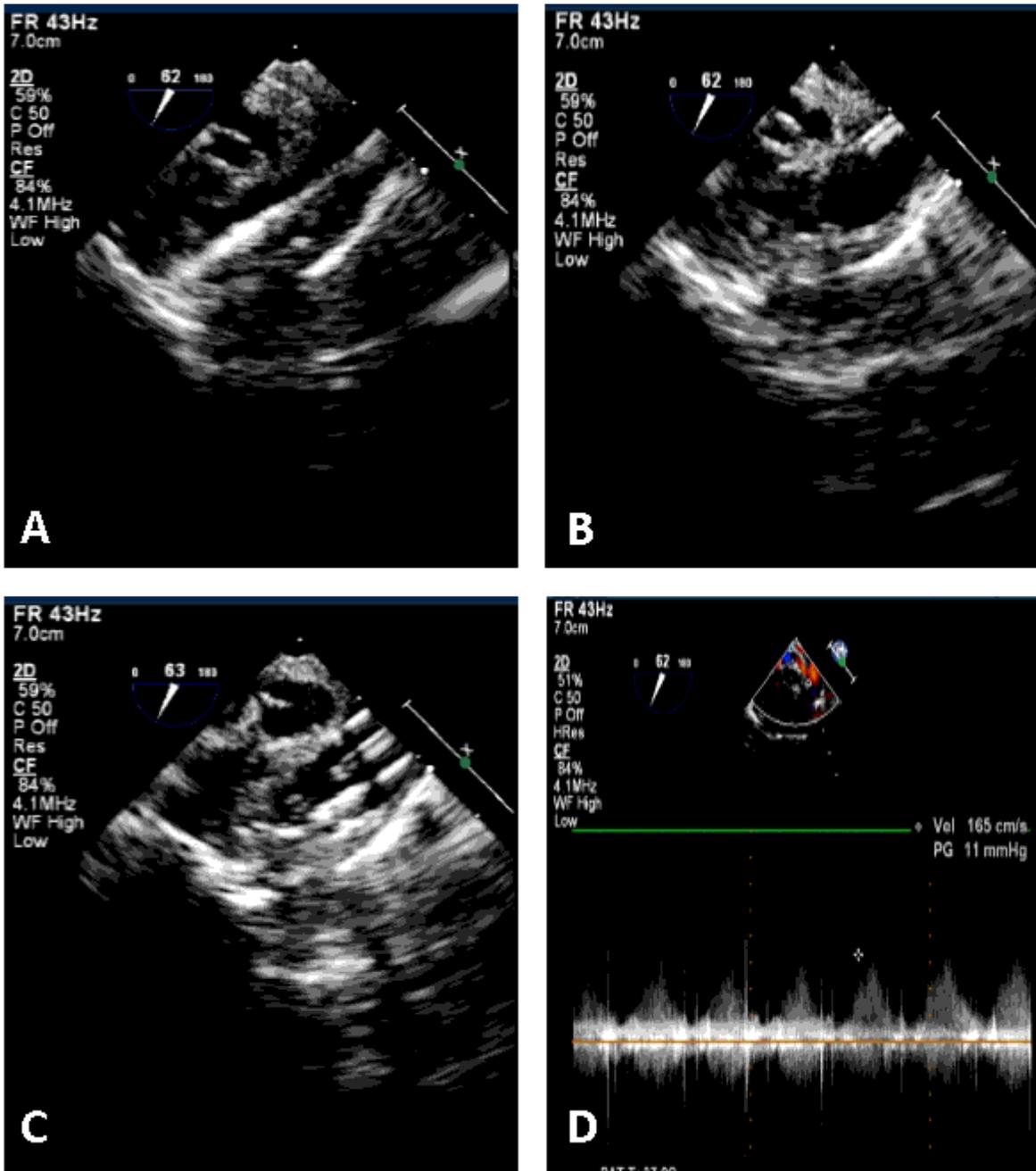
**Figure 2**

3cm incision in the thoracic lower thoracic, open the pericardium and suspension, right coronary artery shown abnormal thickening, diameter 8mm.



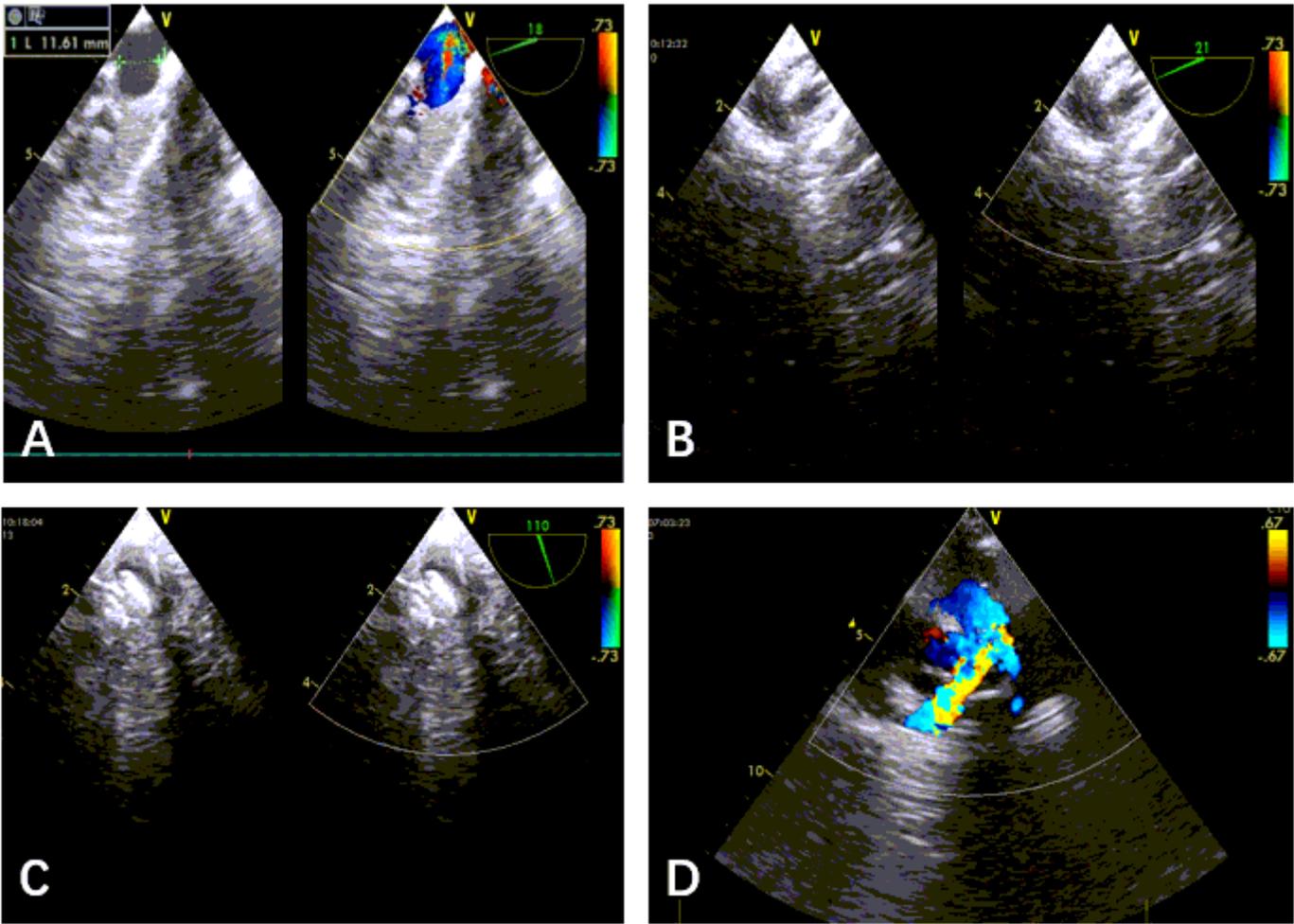
**Figure 3**

A. Right coronary artery trunk diameter 7.6mm, right ventricular opening diameter 5mm, continuous left to right blood flow, the maximum pressure was 56mmHg. B. Right coronary artery - right ventricular fistula after transesophageal echocardiography closure: the umbrellas opened successfully, securely blocked, right coronary artery - right ventricular fistula without residual shunt.



**Figure 4**

Balloon dilation of the pulmonary valve through the right ventricular outflow tract through a thoracic incision guided by TEE. Note: A is the guidewire passing through the pulmonary valve; B is the sheathing tube passing through the pulmonary valve, and it can be seen that the sheathing tube shows the 'dual-track sign' under ultrasound; C is the balloon opening to expand the pulmonary valve; D shows the decrease in the detectable forward flow in the pulmonary artery.



**Figure 5**

Note: A. TEE shows APW, and the measured length of the defect is about 11mm; B. The sheathing tube passes through the APW, and shows the 'double-track sign'. C. Blocking umbrella occludes the APW, and the bypass from left to right disappears; D. There is no significant acceleration of blood flow in the aortic arch and right pulmonary artery.