3D Printing in Medicine

CASE REPORT

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Enhancing management of double outlet right ventricle when the interventricular communication is remote from the arterial roots through three-dimensional printing

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Abstract

Background Double outlet right ventricle with remote interventricular communication presents significant surgical challenges. Traditional imaging often fails to provide the detailed, three-dimensional anatomical insights required for complex cases. Advancements in three-dimensional (3D) printing offer a valuable tool for preoperative planning and decision-making.

Cases In the first case, a 5-year-old with double outlet right ventricle and remote interventricular communication underwent a Glenn procedure with anticipated univentricular repair. 3D printing revealed the potential for enlarging the communication, leading to a one-and-a-half ventricle repair. The second case involved a 2-day-old infant with double outlet right ventricle, aortic arch interruption, and remote communication. At one year, 3D modelling enabled a successful left ventricle-to-aorta baffle.

Conclusion These cases underscore 3D printing's role in improving precision, reducing complications, and potentially lowering costs in managing complex congenital heart disease.

Keywords Child, Double outlet right ventricle/surgery, Printing, Three-dimensional

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Introduction

Double outlet right ventricle describes the situation in which both arterial trunks are predominantly supported by the morphologically right ventricle. Variations in anatomy, including the size and location of the interventricular communication, significantly influence the choice of surgical approach. Biventricular repair is the optimal strategy, although this may not always be feasible [1, 2]. Cross-sectional echocardiography is frequently used for preoperative imaging. When a precise understanding of three-dimensional relationships is required, printing technology can offer significant additional anatomical insights [3]. We describe how printed cardiac models have enhanced our surgical decision-making.

First case

A 5-year-old boy was referred to our center for evaluation of a complex congenital heart defect. Initial neonatal cross-sectional echocardiography revealed double outlet right ventricle and pulmonary atresia, with an interventricular communication remote from the aorta (Fig. 1. The cross-sectional echocardiograms show the location the interventricular communication in relationship with the aorta and the tricuspid valve. The interventricular communication is remote from the aorta with no superior outlet extension, LV: left ventricle, RV: right ventricle). Systemic and pulmonary venous connections were normal, with concordant atrioventricular connections and balanced ventricles of good size. The aorta originated anteriorly from the right ventricle, while the pulmonary trunk was atretic, but with confluent right and left arterial branches. The pulmonary circulation was duct-dependent, so we proceeded successfully to stent the duct.

One year later, after satisfactory cardiac catheterization in preparation for a Glenn procedure, the patient underwent surgical intervention. Intraoperative exploration revealed a medium-sized elliptical interventricular communication beneath the septal leaflet of the tricuspid valve, and hence remote from the aorta. This suggested that routing to the aorta would be challenging. On this basis, we opted for functionally univentricular repair, and performed an atrial septectomy along with a superior cavopulmonary anastomosis.

During the follow-up, cardiac catheterization indicated that the hemodynamic parameters were favourable for completion of the Fontan circulation. We proceeded to cardiac computed tomographic scanning, making a printed model to reassess the potential of re-routing the interventricular communication to the aorta (Fig. 2a, b. Three dimensional modelling showing the view from the right ventricle reveals the location of the remote perimembranous interventricular communication in relation to the orifice of the tricuspid valve and the aortic root). To create the model, we used the cloud-based artificial intelligence-driven segmentation platform created by Axial3D. This technique harnesses deep machine learning along with artificial intelligence, thereby streamlining clinical workflows and enhancing the accuracy of segmentation. Use of annotated medical images, and corresponding ground truth segmentations, provides optimal performance. Careful examination of the model suggested the interventricular communication could safely be enlarged to permit an unobstructed baffle to be placed from the left ventricle to the aorta (Fig. 2c. Planned enlargement of the interventricular communication (purple colour) and the planned suture line of the baffle (yellow line). The interventricular communication is marked with red dashed line).

Hence, we proceeded to create a one-and-a-half ventricle repair. The interventricular communication was enlarged anteriorly and superiorly into the subaortic region (Fig. 3a, b,c. Intraoperative view from the right atrium demonstrating the location perimembranous interventricular communication marked by the surgical forceps. The black silk suture encircles the area that was cut to enlarge the interventricular communication (star) (a & b). View from right ventriculotomy showing the interventricular communication (star) that was enlarged toward the atretic subpulmonary outlet. The cut edges of the enlarged interventricular communication marked in white dashed line (c)). A large patch of bovine pericardium was used to route the communication to the aorta. Continuity between the right ventricle and the pulmonary arteries was then established using a 16 mm

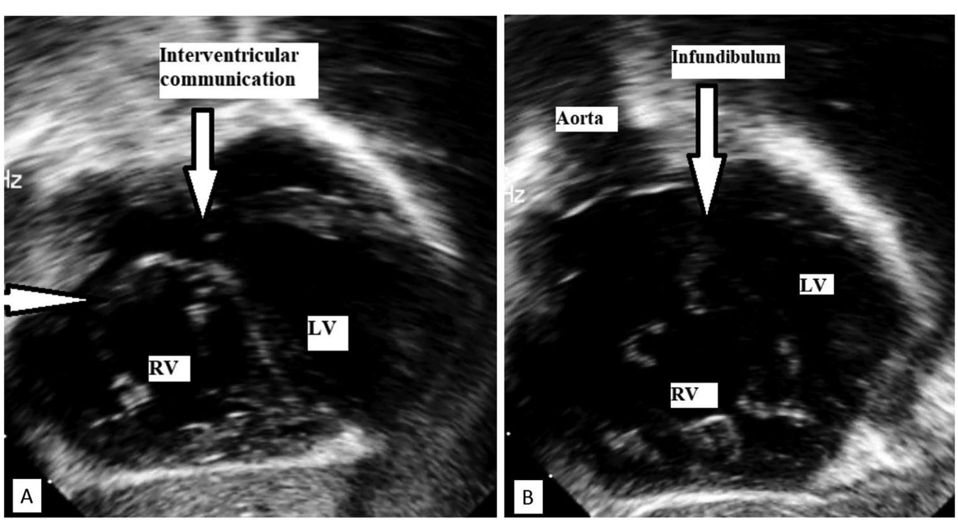

Fig. 1 The cross-sectional echocardiograms show the location the interventricular communication in relationship with the aorta and the tricuspid valve. The interventricular communication is remote from the aorta with no superior outlet extension, LV: left ventricle, RV: right ventricle

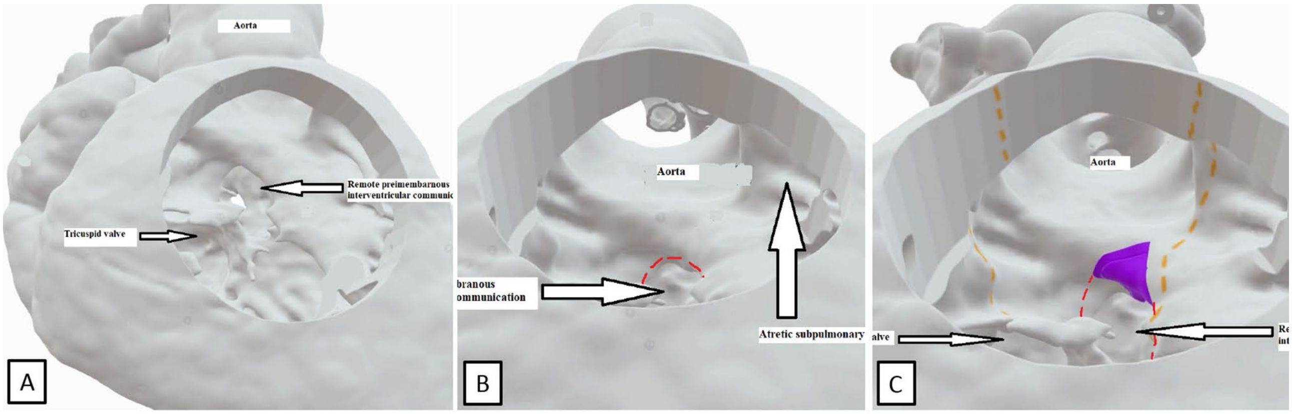

Fig. 2 **a**, **b**,**c**: Three dimensional modelling showing the view from the right ventricle (**a** & **b**) reveals the location of the remote perimembranous interventricular communication in relation to the orifice of the tricuspid valve and the aortic root. Model showing the planned enlargement of the interventricular communication (purple colour) and the planned suture line of the baffle (yellow line and the interventricular communication is marked with red dashed line (**c**)

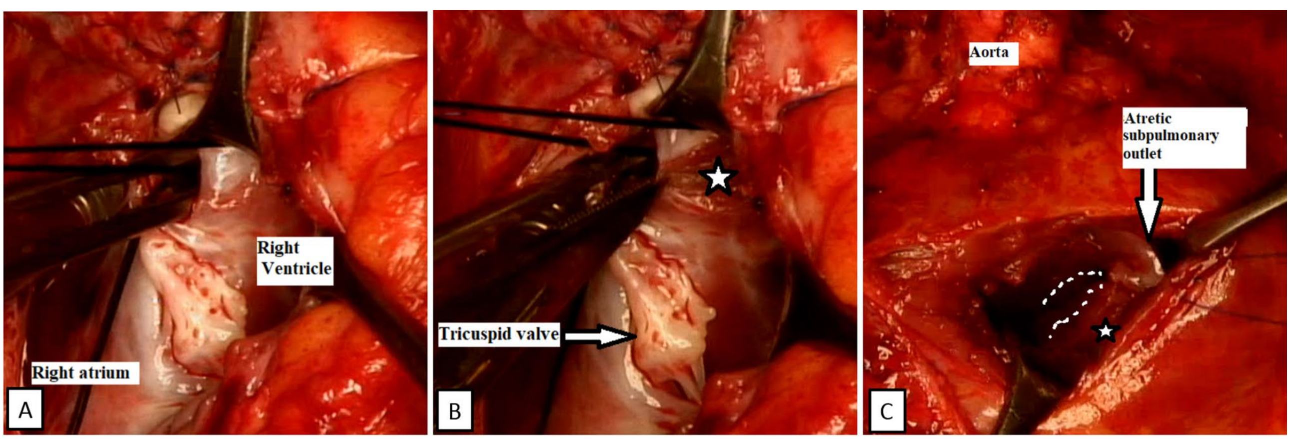

Fig. 3 Intraoperative view from the right atrium demonstrating the location perimembranous interventricular communication marked by the surgical forceps. The black silk suture encircle the area that was cut to enlarge the interventricular communication (star) (**a** &**b**). View from right ventriculotomy showing the interventricular communication (star) that was enlarged toward the atretic subpulmonary outlet. The cut edges of the enlarged interventricular communication marked in white dashed line (**c**)

Contegra® graft, using a cuff of autologous pericardium to augment the pulmonary arterial confluence. We banded the proximal part of the right pulmonary artery to protect the flow from the Glenn shunt from competitive pulsatile flow. Intraoperative transesophageal echocardiogram showed an unobstructed aortic pathway, and good left ventricular systolic function. The systolic pressure in the right ventricle was four-fifths of systemic pressure, with a pressure of 20 millimeters of mercury measured in the non-pulsatile Glenn shunt. The patient was separated from cardiopulmonary bypass and the chest was closed on the next day after surgery.

The patient experienced prolonged mechanical ventilation, with an extended stay in the intensive care unit due to severe biventricular diastolic dysfunction and recurrent pulmonary alveolar hemorrhage. He underwent several bronchoscopy procedures, which were unsuccessful in identifying the origin of the bleeding. We presumed that the bleeding could either be attributed to pulmonary edema resulting from diastolic dysfunction, or be due to the sudden increase of the blood flow in a pulmonary circulation previously dependent solely by the Glenn shunt. Medical therapy, including carvideol, Ivabradine, dapagliflozin, diuretics, and aspirin, was initiated. This served to resolve the pulmonary haemorrhage, permitting eventual extubation after intensive rehabilitation. The patient was discharged after three months of hospitalization, with 96% oxygen saturation. Follow- up after 6 months revealed good biventricular function, mild tricuspid regurgitation with a peak gradient of 45 millimeters of mercury, and an unobstructed pathway from the left ventricle to the aorta (Fig. 4a, b. Pre-operative modelling showed the location of the planned new interventricular communication opening to the subpulmonary outlet tract as seen from the right (a) and left (b) ventricles, along with the planned suture line (yellow

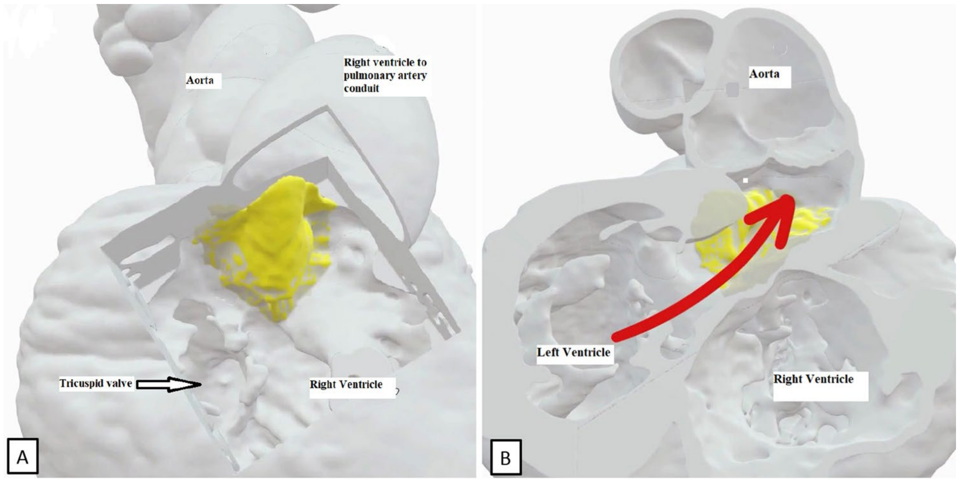

Fig. 4 Postoperative three-dimensional model: View from the right ventricle after a cut in the free wall, highlighting the patch used for the left ventricle-to-aorta baffle and the right ventricle-to-pulmonary artery conduit (**a**). Coronal section demonstrating the direction and configuration of the unobstructed left ventricle-to-aorta baffle (indicated by the red line) (**b**)

lines). Abbreviations: (1) tricuspid valve, (2) the original interventricular communication, (3) the new interventricular communication) (Videoclip-1. The video clip of the three-dimensional modelling of case 1 demonstrates the preoperative anatomy, the surgical planning, and the postoperative anatomy).

Second case

A 2-day-old male infant presented with respiratory distress shortly after birth. Transthoracic echocardiography revealed double outlet right ventricle with interruption of the aortic arch at the isthmus. The scan also revealed presence of a defect across the oval fossa producing a left-to-right shunt, and large but remote interventricular communication (Fig. 5a.Two-dimensional transthoracic echocardiogram). Computed tomographic imaging (Fig. 5b. Computed tomography scan (coronal view) showing the origin of both arterial trunks from the right ventricle and the potential baffle from the left ventricle to the aorta (yellow line). This view illustrates the challenge of creating the baffle without obstructing either the left ventricle to aorta pathway (red line) or the right ventricle-to-pulmonary artery pathway (blue line)) suggested that channelling the left ventricle to the aorta would prove challenging. We proceeded, therefore, to repair the aortic arch via a left thoracotomy, followed by banding of the pulmonary trunk. The postoperative course was complicated, with prolonged mechanical ventilation and a four-month hospital stay. Cardiac catheterization revealed adequate banding, with a gradient of 60 mmHg. Persistent pulmonary over-circulation was noted, likely due to streaming, as indicated by the calculated ratio of pulmonary to systemic blood flows.

Following hospital discharge, the patient was monitored in routine follow-up. At the age of one year, he was referred for definitive surgical intervention. A three-dimensional printed model was obtained (Fig. 5c. Three-dimensional visual model with a cut in the free wall of the right ventricle, showing the tricuspid valve, the location of the interventricular communication, and the positions of the aorta and pulmonary trunk. The presence of bilateral infundibula creates the impression of remoteness of the interventricular communication, but the communication is routable to the aorta, as demonstrated by the proposed yellow suture lines), which demonstrated the location of the interventricular communication. This suggested it would be feasible to create a baffle within the right ventricle to connect the left ventricle with the aorta. The surgery was successfully performed using a bovine pericardial patch (Fig. 6a, b. Postoperative three-dimensional model: View from the right ventricle showing the unobstructed pathway from the right ventricle to the pulmonary trunk, crossing over the left ventricle-to-aorta baffle (yellow patch) (a). View from the left ventricle, illustrating the unobstructed flow and its direction toward the aorta (b)), with smooth postoperative recovery. Follow-up echocardiography revealed no significant residual shunts or obstructions to the arterial outflow tracts. (Videoclip-2. The video clip of the three-dimensional modelling of case 2 demonstrates the preoperative anatomy, the surgical planning, and the postoperative anatomy).

The authors confirm that all methods were conducted in accordance with standard guidelines. The 3D modelling and protocols received approval from the institution's Scientific Research Committee [MOH/CSR/CR/24/6]. Informed consent was obtained from the legal guardians of the children involved.

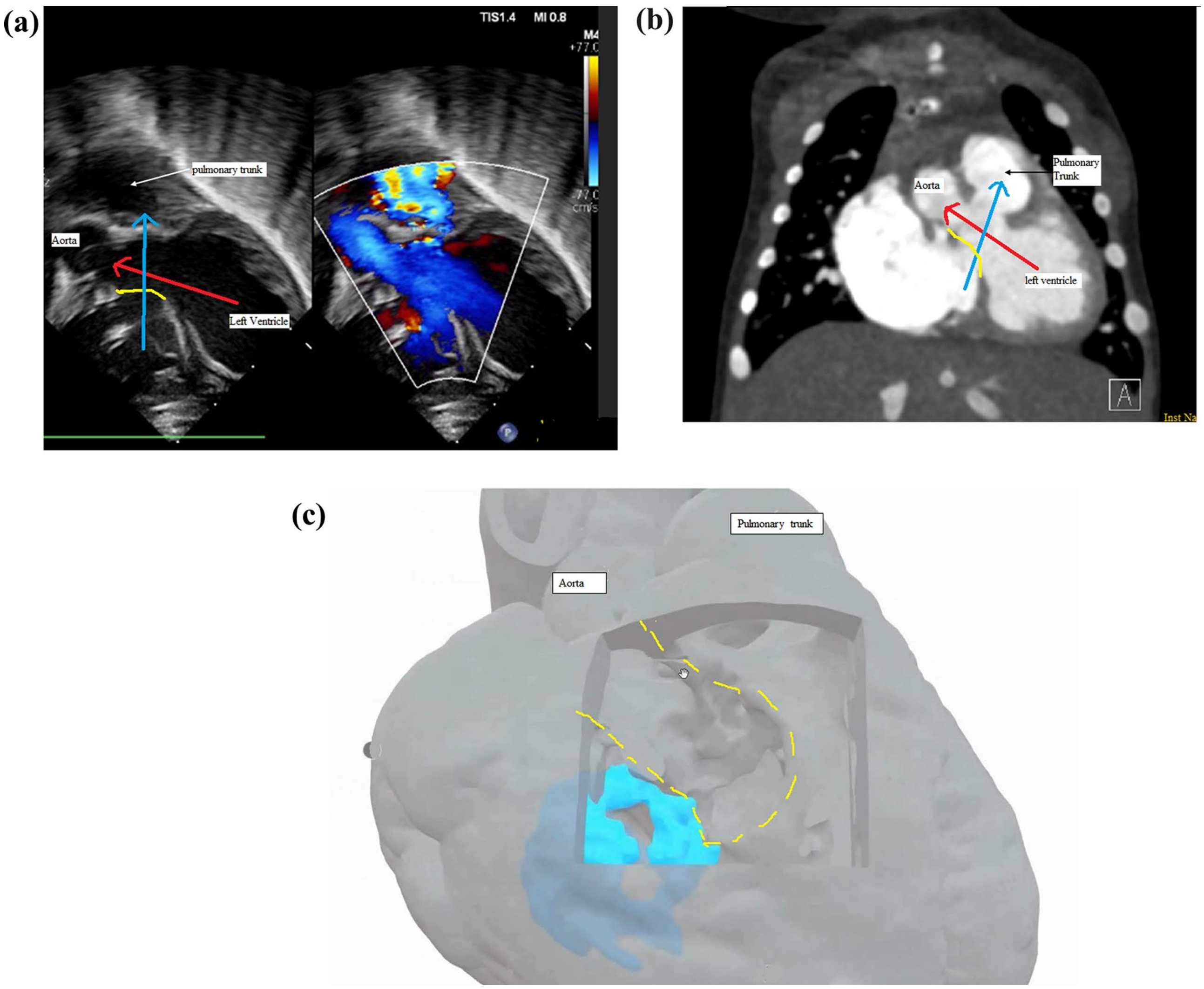

Fig. 5 **a**, **b**,**c**: Preoperative imaging: Two-dimensional transthoracic echocardiogram(**a**) and a computed tomography scan (coronal view) showing the origin of both arterial trunks from the right ventricle and the potential baffle from the left ventricle to the aorta (yellow line). This view illustrates the challenge of creating the baffle without obstructing either the left ventricle to aorta pathway (red line) or the right ventricle-to-pulmonary artery pathway (blue line) (**a**,**b**). Three-dimensional visual model with a cut in the free wall of the right ventricle, showing the tricuspid valve, the location of the interventricular communication, and the positions of the aorta and pulmonary trunk. The presence of bilateral infundibula creates the impression of remoteness of the interventricular communication, but the communication is routable to the aorta, as demonstrated by the proposed yellow suture lines (**c**)

Discussion

Three-dimensional printing has emerged as a transformative tool in the evaluation and management of patients with congenital heart disease. Numerous studies have demonstrated that patient-specific models accurately replicate the complexity of the anatomy, offering significant benefits for preoperative planning and procedural simulations [4]. The technology is particularly valuable in cases where both arterial trunks originate from the morphologically right ventricle [3]. Considerable phenotypic variation is found in this setting, especially regarding the location of the interventricular communication in relation to the great arteries [5], with this feature determining the feasibility of creating an unobstructed baffle from the left ventricle to the aorta [6].

Conventional imaging techniques, while invaluable, often pose challenges in visualizing the intricate cardiac anatomy, especially for less experienced surgeons. These limitations are most apparent when deciding between biventricular and functionally univentricular repairs. Although survival rates for these approaches are comparable, managing long-term complications associated with the Fontan circulation remains a significant challenge [7]. Concerns also persist regarding the exercise capacity of those patients corrected in functionally univentricular fashion [8]. Recent studies have shown no significant survival differences after 10 years between biventricular

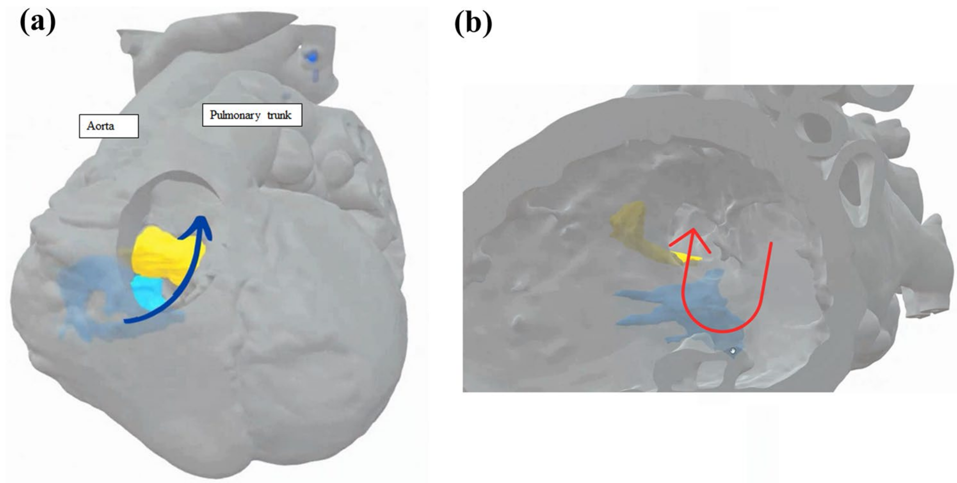

Fig. 6 Postoperative three-dimensional model: View from the right ventricle showing the unobstructed pathway from the right ventricle to the pulmonary trunk, crossing over the left ventricle-to-aorta baffle (yellow patch) (**a**). View from the left ventricle, illustrating the unobstructed flow and its direction toward the aorta (**b**)

and functionally univentricular repairs, though a trend favoring biventricular repair has been noted [9]. Of note, it has been shown that patients initially managed with univentricular palliation can sometimes transition to one-and-a-half or biventricular physiology with favorable outcomes [10].

In our first case, the decision to proceed with a functionally univentricular pathway was influenced by cross-sectional echocardiographic findings. These had suggested the remoteness of the interventricular communication would make a left ventricle-to-aorta baffle difficult to achieve. The creation of a printed model, however, shifted the management strategy toward a one-and-a-half ventricle repair. The model showed with much more accuracy the relationship between the atrioventricular valves and the arterial roots. It also provided critical insights as to how the interventricular communication could be enlarged without compromising structures such as the tricuspid or mitral valves. A further advantage was the site for placement of the right ventriculotomy so as to achieve optimal visualization.

In the second case, in retrospect, had a model had been obtained at the initial presentation, we could have achieved an intracardiac repair at the same time as the reconstruction of the aortic arch. This approach might have optimized our surgical strategy, minimizing the subsequent pulmonary over-circulation, shortening the hospitalization period, and significantly improving the overall clinical outcomes.

After biventricular conversion, patients often face challenges related to "ventricular acclimatization," a term describing the adjustment of the ventricular mass to new hemodynamic conditions [11]. Elevated left ventricular end-diastolic pressures then pose a significant risk for adverse outcomes, including lung congestion, pulmonary edema, prolonged ventilation, and multiple reintubations [12]. A one-and-a-half ventricle repair can serve as an interim step before complete biventricular conversion, avoiding the abrupt preload increase that can stress the systemic ventricle. This staged approach may lead to improved outcomes [10].

The cost of producing the models in our cases was approximately $1,600. When compared with the $70,000 cost of prolonged hospitalization during the second surgery in the first case, it becomes evident that integrating printing of models into standard protocols for managing complex cases could be a cost-effective strategy.

Supplementary Information

The online version contains supplementary material available at https://doi.org/10.1186/s41205-025-00265-y.

Supplementary Material 1: Videoclip-1: The video clip of the three-dimensional modelling of case 1 demonstrates the preoperative anatomy, the surgical planning, and the postoperative anatomy.

Supplementary Material 2: Videoclip-2: The video clip of the three-dimensional modelling of case 2 demonstrates the preoperative anatomy, the surgical planning, and the postoperative anatomy.

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Author contributions
Hamood Al Kindi: conception, design of the work, interpretation of the data, drafted the work Madan Maddali: design of the work and acquisition of the data Pranav Kandachar: helped in the interpretation of data Robert H. Anderson: Interpretation of the data, revised the manuscript.

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Data availability
No datasets were generated or analysed during the current study.

Declarations

Ethical approval
Not applicable.

Consent to Participate
Not applicable.

Consent to Publish
Not applicable.

Informed patient consent
Obtained from parents.

Institutional scientific research committee approval
MOH/CSR/CR/24/6.

Competing interests
The authors declare no competing interests.

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