BRIEF REPORT

Three-Dimensional Printing Model in Double-Outlet Right Ventricle to Simplify Intraventricular Repair

Impresión tridimensional en doble vía de salida del ventrículo derecho para simplificar la reparación intraventricular

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ABSTRACT

Background: Three-dimensional (3D) printing of anatomical structures is gaining interest in congenital heart surgery. We report the case of a 6-year-old boy with double-outlet right ventricle, tetralogy of Fallot subtype, who underwent surgical repair with intracardiac patch. This procedure was performed with the support of two 3D printed models: one of the heart and another of the tunnel patch.
Methods: Transthoracic color Doppler echocardiography, cardiac catheterization, and multi-detector row computed tomography angiography with 3D reconstruction were performed as preoperative evaluation. The 3D models of the heart and the patch –virtually designed to create the intracardiac tunnel– were printed to evaluate possibilities and strategies during the repair. Both 3D models were sterilized for intraoperative use.
Results: The 3D heart accurately showed the position of the ventricular septal defect regarding adjacent structures, including the pulmonary, tricuspid, and aortic valves. The virtual planning, simulation, and the 3D-printed patch model were useful for making the sutured patch for the tunnel. Postoperative color Doppler echocardiography revealed an effective repair with no residual obstruction.
Conclusions: Three-dimensional printing models of the intracardiac anatomy, together with simulation and patch printing, using multi-detector row computed tomography angiography, could provide valuable information for preoperative planning in patients with double-outlet right ventricle, tetralogy of Fallot subtype. The 3D-printed patch models could also be useful to simplify and increase the efficacy of complex procedures.

Key words: Printing, Three-Dimensional - Cardiac Surgical Procedures - Heart Defects, Congenital - Double Outlet Right Ventricle

RESUMEN

Introducción: La impresión tridimensional (3D) de estructuras anatómicas está ganando interés en cirugía de cardiopatías congénitas. Reportamos el caso de un niño de 6 años con doble vía de salida del ventrículo derecho, subtipo Fallot, que fue sometido a reparación quirúrgica con parche intracardíaco. Este procedimiento se realizó con asistencia de dos modelos impresos 3D: uno del corazón y otro del parche para el túnel.
Material y métodos: La evaluación preoperatoria se realizó mediante ecocardiograma Doppler color transtorácico, cateterismo cardíaco y angiotomografía computarizada multidetectores con reconstrucción 3D. Los modelos tridimensionales del corazón y el parche virtualmente diseñados para crear el túnel intracardíaco fueron impresos, para evaluar posibilidades y estrategias en la reparación. Ambos modelos 3D fueron esterilizados para uso intraoperatorio.
Resultados: El corazón 3D mostró en forma precisa la posición de la comunicación interventricular con respecto a las estructuras adyacentes, incluidas las válvulas pulmonar, tricúspide y aorta. La planificación virtual, simulación e impresión 3D del molde del parche fue útil para la confección del parche suturado para el túnel. El ecocardiograma Doppler color posoperatorio evidenció una reparación efectiva sin obstrucción residual.
Conclusiones: Los modelos cardíacos impresos 3D de la anatomía intracardíaca junto con la simulación e impresión de parches, usando angiotomografia computarizada multidetectores, podrían brindar valiosa información para la planificación preoperatoria en pacientes con doble vía de salida de ventrículo derecho tipo Fallot. Los modelos 3D impresos del parche también podrían ser útiles para simplificar e incrementar la eficacia de procedimientos complejos.

Palabras claves: Impresión Tridimensional - Procedimientos Quirúrgicos Cardíacos - Cardiopatías Congénitas - Ventrículo Derecho con Doble Salida

Abbreviations

DORV	Double outlet right ventricle	**CTA**	Computed tomography angiography
VSD	Ventricular septal defect		

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INTRODUCTION

Congenital heart diseases include an extraordinary range of morphological malformations. In-depth understanding of anatomy and physiology is fundamental for diagnosis and management. Imaging modalities including echocardiography, cardiac catheterization, computed tomography angiography (CTA), and magnetic resonance imaging (MRI) aid diagnosis and surgical planning. While the process of image acquisition is often standardized, interpretation can be subjective, depending upon experience, and expertise. (1, 2)

Three-dimensional (3D) printing is an emerging, innovative technology in medicine in constant evolution, that is used in a number of different fields. The interest in pediatric cardiac surgery has increased, and 3D models are able to reproduce complex anomalies of the heart and great arteries. (3) Although the use of three-dimensional models has been reported in different congenital heart diseases, their usefulness in double-outlet right ventricle (DORV) is relevant, as they allow for a more accurate understanding of the complex anatomy, the planning of preoperative strategies, and the possibilities of corrective surgery, improving the outcomes. (3, 4) Double-outlet right ventricle is a type of conotruncal anomaly in which both the pulmonary artery and the aorta originate completely or predominantly in the right ventricle. The international nomenclature of the Society of Thoracic Surgeons and the European Association of Cardiothoracic Surgery defines four DORV subtypes based on the relationship of the ventricular septal defect (VSD) with the great arteries and the presence of right ventricular outflow tract obstruction: 1) DORV VSD-type, (2) DORV Fallot-type, (3) DORV TGA-type (Taussig-Bing), and (4) DORV non-committed VSD. (5, 6)

We report the case of a 6-year-old boy with DORV, tetralogy of Fallot type, who underwent surgical repair with intracardiac patch. This procedure was performed with the support of two 3D-printed models: one of the heart and another of the tunnel patch.

METHODS

A 6-year-old boy, weighing 15 kg, was referred to our hospital due to dyspnea. Physical examination showed 92% oxygen saturation in ambient air, 4/6 systolic murmur in the pulmonary area and decreased pulmonary component of the second heart sound. Blood pressure was 90/60 mmHg, and heart rate was 100 bpm. Color Doppler echocardiogram and cardiac catheterization revealed DORV Fallot-type, with moderate valve and pulmonic subvalvular stenosis (Figure 1). A multi-detector computed tomography angiography (CTA) (Toshiba, Aquilion 64 detectors, Japan) was performed with 3D reconstruction using 0.5 mm slice thickness. It was necessary to reduce the patient's heart rate with a dose of 1 mg/kg oral propranolol 6 h before the study to improve image acquisition. An aberrant right subclavian artery and a coronary anomaly of the anterior descending artery originating in the right coronary artery, crossing the right ventricular outflow tract were found (Figure 2A). The images in DICOM Standard (Digital Imaging and Communications in Medicine) format were sent to an independent work station for the creation of the 3D cardiac model, using open or free software for computer-aided design (CAD), such as InVesalius 3.0 (Sao Paulo, Brazil), Blender 2.76 (Amsterdam, Netherlands), and Meshmixer (California, USA). These softwares made it possible to clean, analyze and process the images initially, and then convert them to a STL (standard tessellation language) file for 3D printing. The heart model was made using a semi-automatic method where a shell (1.5-2 mm thick) was created around the blood pool, with subsequent blood subtraction. Bone structures and unnecessary anatomy were subtracted by traditional methods using the CAD software. The heart model was printed with the Kikai

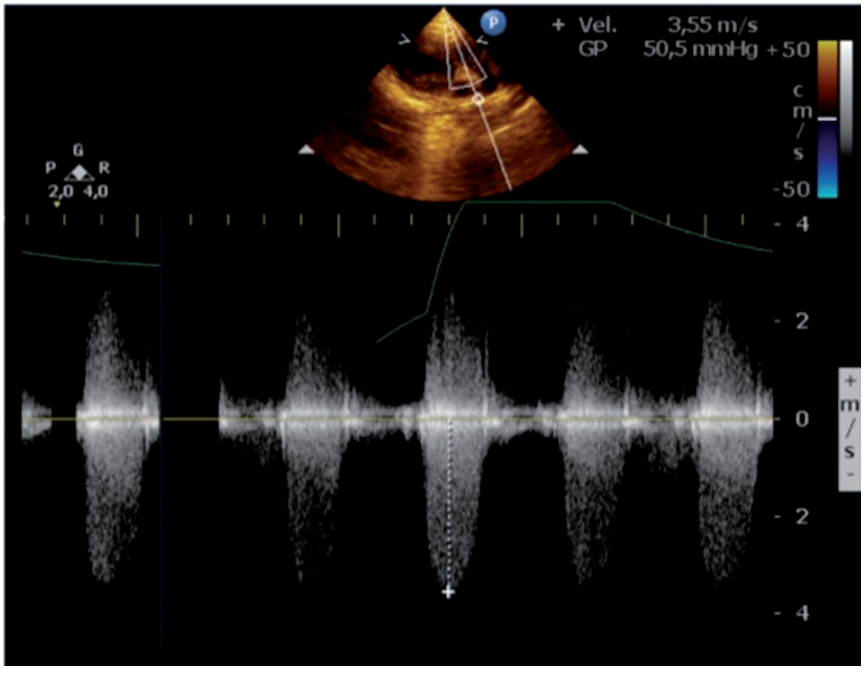

Fig. 1. Preoperative color Doppler echocardiography showing moderate valve and pulmonic subvalvular stenosis.

Labs Maker T125 printer (Buenos Aires, Argentina), which uses thermoplastic polymer extrusion (ABS: acrylonitrile butadiene styrene) and the fusion deposition model (FDM) method (Figure 2B).

The images and the 3D model were evaluated and discussed by members of the multidisciplinary congenital heart disease team and by professionals from the Unit of 3D biomodels. Once the 3D model was printed, the VSD diameter, the distance to the aorta, the possibility of tunneling, the proximity of the tricuspid valve, and the potential size and shape of the patch were evaluated. In addition, the virtual patch for the tunnel was designed and printed for use as a mold during surgery (Figure 3A).

Both 3D models underwent cold gas sterilization for intraoperative use. After median sternotomy, the patient was placed in cardiopulmonary bypass with aortic and bicaval cannulation.

The anterior descending artery emerging from the right coronary artery and crossing the right ventricular outflow tract was visualized, as well as the side-by-side disposition of the great arteries.

The aorta was clamped, cardioplegia was administered, and the right atrium was opened. After retracting the septal tricuspid leaflet, the VSD was visualized. Ninety percent of the aorta was straddling the interventricular septum. The VSD diameter, the distance to the aorta, and the possibility of tunneling were reconsidered and compared with the 3D-printed models of the heart and patch.

The distance from the lower margin of the VSD to the anterior margin of the aortic annulus was measured and compared with the printed patch, which was used as a "mold" to cut the Dacron patch of the tunnel (Figure 3A). Horizontal U- 5-0 prolene stitches were placed on the margin of the VSD, on the muscle margin between the VSD and the tricuspid valve, and around the aortic annulus. Once the patch was inserted through the sutures, it was adjusted and knotted. It was unnecessary to make additional incisions to the patch, since its size was accurate as calculated before surgery (Figure 3B). The right ventricular outflow tract was enlarged with a pericardial patch together with the commissurotomy of the pulmonary valve in order to enlarge and preserve the annulus to a diameter of 15 mm (z score -0.17). After cross-clamp removal, the patient was weaned from cardiopulmonary bypass in sinus rhythm.

RESULTS

The 3D heart accurately showed the position of the VSD regarding adjacent structures, including the pulmonary, tricuspid, and aortic valves. The virtual planning, simulation, and 3D-printed patch model were useful for making the sutured patch for the tunnel. Postoperative color Doppler echocardiography revealed an effective repair with no residual obstruction in the tunnel. A 20 mmHg pulmonary valve gradient was demonstrated, with no failure or residual shunting. The patient was extubated on day 1 after surgery, required antibiotics for an upper respiratory infection, and was discharged on day 14 after surgery. The patient is asymptomatic 16 months after surgery. Color Doppler echocardiography showed good biventricular function, no significant gradient in the right ventricular outflow tract (13 mmHg), mild pulmonary valve regurgitation, and no tunnel obstruction.

DISCUSSION

Three-dimensional printing technology is used to create solid objects in three dimensions from digital

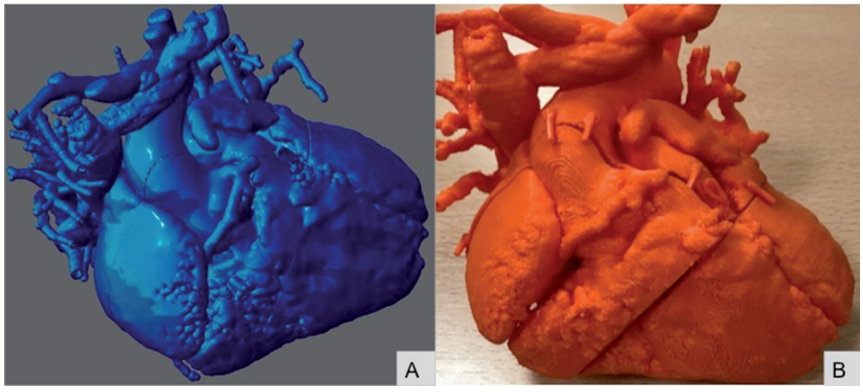

Fig. 2. A: Anterior 3D reconstructed virtual image of the blood pool used to create the 3D printing. **B:** Anterior view of the 3D-printed model with the fusion deposition model (FDM) method, printed in 4 segments for intracardiac access

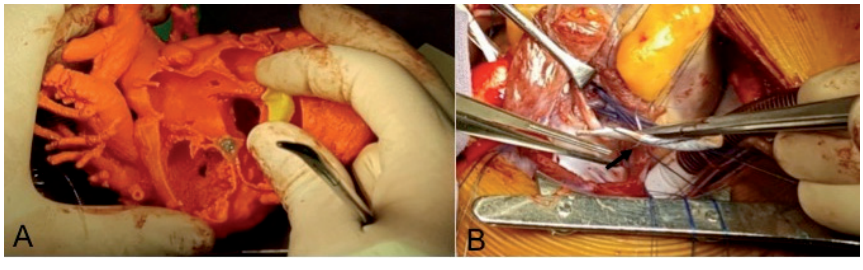

Fig. 3. Intraoperative comparison between 3D model (A) and patient anatomy. Intracardiac patch suture to create the tunnel (B).

images. (7) As recently reported, this technology has been used for the diagnostic optimization and surgical planning of different congenital heart diseases, such as VSD, vascular annulus, univentricular hearts, pulmonary venous return anomalies, hypoplastic left heart syndrome, tetralogy of Fallot, DORV, and complex variants of transposition of the great arteries. (3, 4, 8-15) However, it is in the surgical planning of patients with DORV that 3D cardiac models may be most useful, as published by Farooki K et al. (8) and Garekar et al, (9) respectively, as it allows understanding the relationship of VSD, great arteries and valves to determine and design the shape, size and orientation of the intraventricular tunnel, while visualizing, at the same time, anatomical intracardiac relationships. (10, 16)

Double outlet right ventricle morphology is affected by a careful description of VSD and its association with the sigmoid valves, great vessels, coronary anatomy, the presence of right and left ventricular outflow tract obstruction, the distance between the pulmonary and the tricuspid annulus, and the presence or absence of associated cardiac lesions. (6) The treatment of choice in DORV Fallot-type patients includes intraventricular tunnel repair and release of right ventricular outflow tract obstruction, as performed in the present report. (12)

In our case, preoperative planning was simplified with 3D printing of the heart and patch. Ventricular septal defect visualization and its relationship with the aorta in the printed model was similar to the surgical observation. Dimensions and structures in the 1:1 scale models were similar to those of the patient. A large portion of the ventricular septum was clearly observed, as well as VSD, aortic annulus, pulmonary annulus, and also the coronary anomaly crossing the right ventricular outflow tract. This technology allowed us to plan the path and size of the tunnel and a safe infundibulotomy.

We agree with Hibino N. et al, (12) that computer simulation with CAD would facilitate the creation of patient-specific implants. The accuracy of the intraventricular tunnel model was optimized with the technique used. We believe that printing the virtually designed 3D patch and using it as a mold is very useful. Like Farooqi K. et al., (8) we demonstrated that 3D printing offers the unique chance for the surgeon to visualize possible tunnel paths in three dimensions. Sodian R. et al. (13) published that being able to hold a model in one's hand and examine it from different sides optimizes the surgical approach and allows anticipation of possible problems. Dimensions and distances can be identified, as in our case. We do not believe that these 3D models are necessary in all pediatric cases, but they allow the surgeon to better understand patient-specific three-dimensional anatomy in complex heart diseases such as DORV Fallot-type, improving preoperative planning and intraventricular orientation of the tunnel, and thus optimizing the outcomes. A multidisciplinary cooperative approach between radiologist, surgeon, cardiologist and computer and design specialists is essential to achieve a precise 3D model. (13)

In conclusion, this innovative method could be useful for surgical planning and execution of complex congenital heart diseases such as DORV, as it optimizes the understanding of the three-dimensional anatomy and allows to anticipate the technical challenges of the pathology. Three-dimensional models of other complex heart diseases will be needed to further evaluate the usefulness of this technology.

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Conflicts of interest

None declared. (See authors' conflicts of interest forms on the website/Supplementary material).

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