

Research Article

3D-Printed Models for Multidisciplinary Discussion of Congenital Heart Diseases

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Background. Congenital heart defects (CHDs) are complex three-dimensional (3D) lesions with variable anatomies that present therapeutic challenges. The application of a patient-specific3D-printed model in preoperative planning and communication in medical practice can contribute to a complete understanding of the intracardiac and vascular anatomy. This study aimed to prospectively investigate the clinical value of a 3D CHD model in multidisciplinary discussions. Methods. Between August 2019 and April 2021, 19 patients with complex CHDs before surgery were prospectively enrolled in this study. Eight to 14 medical specialists participated in multidisciplinary discussions using patient-specific 3D models. A subjective satisfaction questionnaire, comprising 12 questions to be answered on a 10-point scale, was distributed. Results. Twenty 3D-printed anatomic models of 19 patients were used. The median age and weight of the enrolled patients were 0.8 years (range, 5 days to 43 years) and 9.6 kg (range, 2.8-54 kg), respectively. The most common underlying disease was a double outlet of the right ventricle. The mean scores for understanding spatial orientation, ease of communication between clinicians during discussions, prediction of surgical complications, and information additional to conventional 2D imaging were 9.4 ± 1.1 , 9.4 ± 0.9 , 9.0 ± 1.1 , and 9.2 ± 0.4 , respectively. The competency and comfort scores for each patient's surgical plan increased significantly after using the 3D-printed model (from 6.2 ± 1.6 to 9.2 ± 0.9 , p < 0.001 and from 6.3 ± 1.6 to 9.2 ± 0.8 , p < 0.001, respectively). Conclusions. Patient-specific 3D models, for patients with complex CHDs, improved the understanding of the disease and facilitated multidisciplinary discussions and surgical decision-making. However, because outcomes were mainly evaluated by subjective reports, the possibility of other unknown factors affecting the outcomes should be considered. Trial Registration. This trial is registered with D-1904-031-1024.

1. Introduction

Congenital heart defects (CHDs) are complex and widely variable anatomic lesions that present serious diagnostic and therapeutic challenges. Three-dimensional (3D) printing allows understanding of the 3D orientation and spatial relationship of cardiovascular structures in CHDs. 3D-printed anatomic models have had various applications in trainee education, surgical/interventional planning, patient/family education, and communication in medical practice [1–4]. However, due to

their challenging nature, there are still knowledge gaps and limited data in this area in terms of randomized studies and comparative research on the outcomes of 3D printing, especially considering the variety of available software, hardware, techniques, and printing materials [5]. Different centers have reported different experiences and practices in 3D printing. Therefore, we prospectively investigated the clinical value and feasibility of a 3D-printedpatient-specific model for multidisciplinary discussions of various complex CHDs in a single tertiary center.



FIGURE 1: Construction of the three-dimensionally-printed model.

2. Materials and Methods

From August 2019 to April 2021, we conducted an openlabel prospective pilot study, using 20 3D-printed models of 19 patients with complex CHDs during 20 multidisciplinary discussions among 8-14 pediatric cardiologists and cardiothoracic surgeons. The selection of cases for 3D printing was decided at a multidisciplinary meeting. After obtaining informed consent from the patients and/or their parents, depending on the participant's age, a cardiac computed tomography (CT) scan was used to generate a DICOM file. Segmentation and postprocessing of the cardiovascular structure were performed, and standard tessellation language files were generated using commercially available software (MEDIP PRO v2.0.0., MEDICAL IP, Seoul, Korea) (Figure 1). Patient-specific3D-printed models were produced in two types: "blood pool model" and "hollow model," which consisted of the lining around the blood pool model with a wall thickness of 1-2 mm, meticulously representing the intracardiac anatomy (Figure 1, right bottom) [6]. Using the 3D-printed model, we discussed the management plan in a multidisciplinary meeting. All participating cardiologists and cardiac surgeons were given a questionnaire, to which they had to respond on a scale of 0–10, with 10 indicating the highest score. The questions in the questionnaire surveyed the effect of the model in understanding the 3D orientation of the cardiovascular anatomy, designing a surgical plan, predicting surgical complications, facilitating multidisciplinary discussions, and communication, how it changes comfort and confidence in management, and if there was a change in the management plan after the use of the 3D-printed model (Supplementary Table 1). Patient demographic and clinical data were extracted from electronic medical records.

An intraclass correlation coefficient and Bland–Altman plot were obtained for 20 printed models to illustrate the agreement between the phantom CT of the 3D-printed model and the cardiac CT (Supplementary Figure 1). The sizes of the ventricular septal defect (VSD), aorta, superior vena cava, inferior vena cava, and pulmonary arteries were measured and compared between the 3D-printed model and phantom CT.

The study was approved by the Institutional Review Board of the Seoul National University Hospital (No. 1904-031-1024). Written informed consent for participation from the patients and/or their parents was obtained before producing the 3D model. Moreover, informed consent was obtained from the clinicians who participated in the discussion and answered the questionnaire.

3. Statistical Analysis

Continuous and ordinal variables were expressed as means \pm standard deviations or medians and ranges, as appropriate. Pre- and post-3D model confidence and comfort scores were compared using the Wilcoxon signed-rank test. Categorical variables are expressed as frequencies (percentages). The chi-square and Fisher's exact tests were used to compare categorical data between two or three groups. Statistical analyses were performed using SPSS software (version 23.0, IBM Corporation, Armonk, NY, USA). Statistical significance was established at p < 0.05.

4. Results

During 20 multidisciplinary discussions, a total of 212 questionnaires were completed. The median age and weight of the patients for whom 3D models were printed were 0.8 years (range, 5 days to 43 years) and 9.6 kg (range, 2.8 to 54 kg), respectively. The clinical diagnosis and reason for printing a 3D model for each patient are summarized in Table 1. There were 12 patients with a double outlet of the right ventricle (DORV), which was the most common underlying disease. In those patients, the 3D models were used in the discussion that led to the selection of the treatment plan between biventricular repairs versus Fontan palliation. Three patients had multiple or unusual locations of VSDs. In two patients with hypoplastic left heart disease and bilateral

1 43y 54 Complete TGA, VSD, FS, subactic stenosis VSD location for VSD widening without heart block Successful vSD ex- heart block 2 4m 44 PA VSD MAPCA Spatial relationship of MAPCA, pulmonary artery Successful viol 3 12m 8.8 TOF, subactic VSD, repaired TAPVR, PA banding Vanual VSD location Total con 4 4y 12 DORV, subacric VSD, repaired TAPVR, PA banding Unusual VSD location Total con 5 27m 10 Muscular VSD, repaired TAPVR, PA banding VSD location Total con 7 5d 3.5 PA, VSD, crise-cross heart Simulation before Norwood operation Total con 7 5d 3.5 PA, VSD, crise-cross heart Simulation before Norwood operation Successful VSD 9 3m 46 HLHS, PAB state Simulation before Norwood operation Successful VSD 11 3m 12 Left isomerism, DORV, caVSD, PS, multiple muscular Simulation before Norwood operation Successful VSD 11 3m 12 DORV, superior ventricle Feasibility for	Model number	Age	Weight (kg)	Diagnosis	Reason for 3D model	Result
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	septal defect		/			(and the farmer of farming (and the

TABLE 1: Patients' diagnosis and reasons for 3D-printed model.

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pulmonary artery banding, 3D models were generated for the simulation of a Norwood operation and in one patient with pulmonary atresia and major aortopulmonary collateral arteries (MAPCAs), a 3D model was created for the unifocalization plan.

4.1. Two Cases as Examples of Surgical Decision-Making Using 3D-Printed Models. A 43-year-old female patient (Table 1, model 1) with complete transposition of the great arteries, a VSD, and pulmonary stenosis had severe subaortic stenosis (pressure gradient between the aorta and left ventricle = 73 mmHg, left ventricular pressure = 196 mmHg) due to a restrictive VSD after a Rastelli operation at the age of 13 years. Her NYHA functional class was II-III, and the stenosis had gradually progressed. Widening of the VSD was required, but there was a high risk of heart block. We had multiple discussions with pediatric cardiologists, cardiothoracic surgeons, radiologists, and pathologists, using a 3Dprinted model, on how to extend the VSD without causing heart block. Eventually, the patient underwent subaortic muscle resection without complications. A transaortic approach and posterior muscle resection were performed to avoid conduction bundle injury. The subaortic stenosis was relieved, and the NYHA class improved to I-II (Figure 2).

A neonatal male patient (Table 1, models 6 and 13) with a superoinferior ventricle, DORV, subaortic VSD, rightsided atrial appendage juxtaposition, a nearly single atrium, and mesocardia in situs solitus (Videos 1 and 2 and Figure 3). The VSD plane was not fully understood via CT and echocardiography. Biventricular repair was initially impossible because VSD baffling would likely cause subpulmonic stenosis, and there was no space for conduit placement between the right ventricle and the pulmonary artery. Although the patient had progressive congestion and mild cardiomegaly, we decided to closely monitor him and wait for biventricular repair. The pulmonary artery had grown with age, and at 11 months of age, the 3D-printed model suggested a possibility of VSD baffling without risking subpulmonic obstruction. Consequently, biventricular repair with VSD baffling, ASD patch partitioning, and right ventricular outflow tract widening was performed.

A good correlation was observed between chest CT and phantom CT, with an intraclass correlation coefficient of 0.996 (95% clinically important difference, 0.993–0.997, p < 0.001) and a mean difference of 0.195 ± 0.681 mm. The Bland–Altman plot revealed no significant bias (Supplementary Figure 2).

4.2. Multidisciplinary Discussion. According to the answers to the questionnaire, the 3D-printed model accurately represented cardiac structures (9.4 ± 0.7) , helped clinicians understand spatial orientation (mean rating score, 9.4 ± 1.1), allowed for easy and quick communication among coworkers $(9.4 \pm 0.9$ and 9.2 ± 1.1 , respectively), aided in the prediction of surgical complications (9.0 ± 1.1) , and provided additional information over conventional imaging (9.2 ± 0.4) (Supplementary Figure 3). Comfort and confidence in the surgical plan significantly increased after using

the 3D-printed model (pre, 6.2 ± 1.6 versus post, 9.2 ± 0.9 , p < 0.001 and pre, 6.3 ± 1.6 versus post, 9.2 ± 0.8 , p < 0.001, respectively) (Figure 4). Pediatric cardiologists and cardiac surgeons did not differ significantly in whether 3D-printed models accurately displayed the cardiac structure, helped to understand the 3D orientation, or simplified communication between clinicians, or in their preoperative /postoperative comfort and confidence after using a 3D-printed model. However, they differed regarding whether the model shortened the discussion; the median rating score awarded by cardiologists and cardiac surgeons were 9.37 ± 0.87 and 9.03 ± 1.32 , respectively (p = 0.033). When prompted to provide additional comments, respondents mentioned limitations, such as that the 3D-printed model did not satisfactorily represent the valve, and that the simulation was inaccurate because the material with which the 3D model was printed differed from actual heart tissue.

5. Discussion

Our study demonstrated that patient-specific3D-printed models accurately represented the cardiac structure, except for the cardiac valve, exhibiting good correlation between chest CT and phantom CT of the models (intraclass correlation coefficient of 0.996 and no significant bias according to the Bland-Altman plot). They also provided insight into the 3D spatial orientation of the defects and helped physicians in their decision-making on the surgical plan and in the prediction of surgical complications. The 3D model facilitated communication and reaching an agreement in multidisciplinary discussions. Comfort and confidence in the surgical plan increased significantly with the 3D model, which illustrates the importance of this tool in preparation for surgery. Furthermore, pediatric cardiologists and cardiac surgeons did not differ in the degree to which they felt the models facilitated decision-making, communication, and their understanding of the 3D anatomic orientation of the defects. Although they differed in their opinions regarding the degree to which the models shortened discussions, both mean scores were more than 9.0 $(9.37 \pm 0.87 \text{ and } 9.03 \pm 1.32, \text{ respectively})$. Taken together, both pediatric cardiologists and cardiac surgeons found the 3D models helpful in various ways, and cardiologists particularly thought that the 3D model shortened the discussion on patient management for CHDs.

Randomized trials or comparative studies on patients with complex CHDs are challenging to perform because they are rare and extremely heterogeneous. Patients differ in the anatomy, combinations of VSD locations, relationship between the great vessels and ventricles, and sizes of the ventricles, VSDs, atria, great arteries, and cardiac chambers. Although difficult to quantify, a patient-specific3D-printed cardiovascular model and surgical/interventional simulation may decrease pump and procedure times, decrease complications, and improve surgical/interventional outcomes.

3D-printed models have been used in patients with CHDs since the early 2000s, and patient-specific 3D models have become more widely used in surgical planning, surgical/percutaneous interventional simulation, and patient/



FIGURE 2: A 43-year-old patient with severe subaortic stenosis. This patient had complete transposition of the great arteries, a VSD, and pulmonary stenosis, and underwent VSD baffling and a Rastelli operation at 13 years of age. However, the previous VSD became restrictive, and the subaortic stenosis gradually progressed. (a) Left ventricular angiography revealing subaortic stenosis. (b) Upon echocardiography in the high parasternal modified long-axis view, the peak velocity at the subaortic level was measured as 5 m/s. (c) The left lateral cutting plane view after 3D segmentation using the software. The white arrow indicates the expected location of the conduction system. (d) The patient-specific3D-printed hollow model. The thick arrow indicates the restrictive VSD. VSD widening was planned using a transaortic approach (arrow). LV, left ventricle; VSD, ventricular septal defect.

family education and communications in the last decade [5]. Patient-specific 3D models have exhibited good agreement with CT scans and magnetic resonance images, while the technique and its application have advanced and evolved into augmented and virtual reality [2, 6]. Models help in surgical decision-making but can also lead to changes in the surgical plan [7, 8]. For example, cross-sectional images alone are insufficient for assessment of the feasibility of biventricular repairs by VSD baffling, as the intracardiac space is limited, and the conduit is positioned between the right ventricle and pulmonary artery, resulting in obstruction. This is especially true in patients with a DORV and remote or subpulmonic VSDs, and in those with an unusual superoinferior relationship of the ventricles. However, the 3D model allows clinicians to visualize the anatomical

relationship between cardiovascular structures, which may lead to a modification of the surgical method to be used [8, 9]. Decision-making regarding Fontan surgery versus biventricular repair requires meticulous consideration, particularly in patients who are suboptimal candidates for biventricular repair, as the decision directly impacts the patient's long-term prognosis. The usefulness of 3D anatomic models in such complex and controversial decisionmaking is indisputable [10].

In our study, the 3D-printed model also helped the surgical team to select a surgical method and allowed simulation in a challenging case that required VSD extension after a Rastelli operation for complete transposition of the great arteries, a VSD, and pulmonary stenosis (Figure 2). In patients with pulmonary atresia and MAPCAs, the 3D



FIGURE 3: A neonatal patient with a superoinferior ventricle, DORV, subaortic VSD, right-sided juxtaposition of the atrial appendages, and mesocardia in situs solitus. The main pulmonary trunk was located posteroinferiorly, the subvalvular infundibulum exhibited mild to moderate stenosis, and the main and branch pulmonary arteries were elongated. The entire heart was rotated clockwise and inferiorly along the axis from the apex to the base. The LV apex was pointing in the subxiphoid direction, where apical beating was visible (a, d). The 3D-printed model clearly showed the spatial relationship of the sternum, ventricle, atrium, and great vessels, as well as the intracardiac ventriculoarterial relationship in this complex heart disease (b, c, e, f). Ao, aorta; AP, anteroposterior; DORV, double outlet of the right ventricle; IVC, inferior vena cava; LAA, left atrial appendage; LV, left ventricle; MPA, main pulmonary artery; PA, pulmonary artery; RAA, right atrial appendage; RV, right ventricle; RVOT, RV outflow tract; TV, tricuspid valve; VSD, ventricular septal defect.

model helped clarify the anatomic and spatial relationship of the MAPCAs, native pulmonary artery, and airways.

The use and applications of 3D modeling and printing have increased and are constantly evolving. It has advanced to the point where it is being used in hands-on surgical training and in computer-aided sterilizable templates for baffles and patches for complex surgical procedures [10]. However, its application and utility differ greatly from country to country and institution to institution, which may be because of differing patient groups and financial/insurance contexts. The process of 3D printing is time-consuming, laborintensive, and expensive, which are obstacles to using 3D models. In our experience, creating a 3D model takes at least seven days, as the segmentation requires manual editing by CHD medical experts, communication between medical experts and software experts, and further refinement with computer-aided design software. Furthermore, the valve and subvalvular apparatus cannot currently be accurately reproduced with a 3D-printed model. The texture of the model differs from that of the real heart, and the models are more easily cut or torn than real cardiac tissue. Although



FIGURE 4: Questionnaire satisfaction survey for the change in comfort and confidence in surgical management and confidence before and after using the 3D-printed model. Both comfort in deciding the management plan (a) and confidence in the management plan (b) significantly increased after using the 3D model (pre, 6.2 ± 1.6 versus post, 9.2 ± 0 , p < 0.001 and pre, 6.3 ± 1.6 versus post, 9.2 ± 0.8 , p < 0.001, respectively).

software, hardware, and 3D-printing material have improved during the study period, further study and improvements are required.

6. Conclusion

Patient-specific3D-printed models improved the understanding of complex CHDs and facilitated multidisciplinary discussions and surgical decision-making in our study. However, a limitation of this study is that the measured outcomes were based on subjective reports. Their wider use will require further study and improvements in terms of the cost and time for necessary for their production, as well as the materials from which they are constructed.

Data Availability

The data that support the findings of this study are available from the corresponding author upon reasonable request.

Ethical Approval

The study was approved by the Institutional Review Board of the Seoul National University Hospital (No. 1904-031-1024).

Consent

Written informed consent for participation from the patients and/or their parents was obtained before producing the 3D model. Moreover, informed consent was obtained from the clinicians who participated in the discussion and answered the questionnaire.

Conflicts of Interest

Soon Ho Yoon works in MEDICAL IP and holds stock of the firm. The other authors declare that they have no conflicts of interest.

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Supplementary Materials

Supplementary Table 1. Questionnaire for Patient specific 3D-printing model. Supplementary Figure 1. Phantom CT of the 3D model 2. Phantom CT was performed for twenty 3Dprinted models. The sizes of the ventricular septal defect (VSD), aorta, superior vena cava, inferior vena cava, and pulmonary arteries were measured and compared between the 3D-printed model and phantom CT. Supplementary Figure 2. Bland-Altman plot for the 3D model and phantom model. Bland-Altman plot for intermeasurement agreement. Limits of agreement are shown as dotted lines with 95% confidence intervals and regression fit of the differences on the means (as solid line). Supplementary Figure 3. Result of Questionnaire satisfactory survey. Video 1. Echocardiography of model 6 shows the superior right ventricle, tricuspid valve, and inferiorly located LV. Both the aorta and pulmonary trunk originate from the right ventricle. Video 2. An anterior oblique view on echocardiography of model 6 shows the left pulmonary artery with stenosis. (Supplementary Materials)

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