Utilisation of three-dimensional printed heart models for operative planning of complex congenital heart defects

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Abstract

Background and aim: To evaluate the accuracy of the three-dimensional (3D) printing of cardiovascular structures. To explore whether utilisation of 3D printed heart replicas can improve surgical and catheter interventional planning in patients with complex congenital heart defects.

Methods: Between December 2014 and November 2015 we fabricated eight cardiovascular models based on computed tomography data in patients with complex spatial anatomical relationships of cardiovascular structures. A Bland-Altman analysis was used to assess the accuracy of 3D printing by comparing dimension measurements at analogous anatomical locations between the printed models and digital imagery data, as well as between printed models and in vivo surgical findings. The contribution of 3D printed heart models for perioperative planning improvement was evaluated in the four most representative patients.

Results: Bland-Altman analysis confirmed the high accuracy of 3D cardiovascular printing. Each printed model offered an improved spatial anatomical orientation of cardiovascular structures.

Conclusions: Current 3D printers can produce authentic copies of patients` cardiovascular systems from computed tomography data. The use of 3D printed models can facilitate surgical or catheter interventional procedures in patients with complex congenital heart defects due to better preoperative planning and intraoperative orientation.

Key words: three-dimensional printing, heart models, congenital heart defects

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INTRODUCTION

A precise evaluation of congenital heart defects anatomy is essential for optimal surgical planning. Standard imaging modalities such as echocardiography, catheter angiography, computed tomography (CT), or magnetic resonance imaging (MRI) are routinely used in this indication. However, these standard methods might not be sufficient for the exact imaging of three-dimensional (3D) cardiovascular anatomical relationships in complex congenital heart defects. The methods mentioned above can provide 3D virtual reconstructions, but only 3D printed heart models based on CT or MRI data can provide physicians with real 3D tactile replicas of complex cardiac anatomies with the benefit of detailed visualisation from all possible perspectives [1]. This invaluable information is extremely helpful for surgical decision-making because even minor anatomical details can indicate the optimal surgical approach in each individual patient with a complex congenital heart defect. Hence, the use of 3D heart models allows one to avoid unexpected findings, especially in very rare congenital heart defects [2]. Thanks to these benefits, 3D model printing has nowadays become an established complementary imaging technique in paediatric cardiology [3, 4]. In the paper pre-

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sented herein, we report our single-centre experience with the initial use of 3D heart model printing in the perioperative planning of patients with complex congenital heart defects.

METHODS

Between December 2014 and November 2015, eight patients (age range 3 days - 30 months) with complex congenital cardiovascular anomalies with ambiguous spatial anatomical relationships were selected for the study. Fabrication of all models was indicated by a cardiac surgeon with the intention of obtaining the most realistic image of complex cardiac defects in patients with discrepancies in their echocardiographic, angiographic, and CT findings. Initially, precise 3D digital information of cardiovascular structures was obtained from CT angiography (Philips Brilliance 40, Amsterdam, Netherlands) in all infants. The image data were stored in DICOM format. An open source software package "3D slicer 4.3" for visualisation and medical image computing was used for DICOM data processing [5]. Firstly, grey-scale manipulation increased the contrast boundaries between the blood pool and myocardium/vessel walls. The segmentation process continued with the extraction of the region of interest (ROI) from the surrounding tissues, i.e. the extraction of hyperdense cardiovascular lumens from hypodense walls of vessels and cardiac chambers. After the ROI extraction, the model surface was smoothed and model data were triangulated and stored in stereolithographic file format (.stl). These data were easily recognised by either the 3D printing device "FELIX 3.0" (FELIXrobotics, De Meern, Netherlands) in the first four patients, or "Zortrax M200" (Zortrax S.A., Olsztyn, Poland) in the consecutive four patients. The additive technique of fusion deposition modelling was employed for the life-like, 1:1, 3D cardiac model printing. Printing filaments "Felix PLA red" compatible with "FELIX 3.0" printer and "Z-ABS" compatible with "Zortrax M200" printer were selected for the models' fabrication. The width of one layer in vertical scale was 0.1 mm, which provided an ideal ratio between both the velocity and spatial accuracy of printing. Once the 3D model printing was done, supporting material was removed manually.

"Cast" models, representing the outside borders of cardiovascular lumens, were fabricated for the exact spatial evaluation of aortic arch, pulmonary arteries, aortopulmonary collaterals, and pulmonary venous drainage anomalies. Moreover, one model was used for the imaging of ventricles, ventricular septal defect, and its spatial relation to great vessels in one patient.

According to the size of the model, the duration of segmentation and printing process lasted between 6 and 12 h. The average cost of the presented models was 300 Euros per model. We fabricated our 3D printed models in collaboration with the Institute of Simulation and Virtual Medical Education of Comenius University, Faculty of Medicine Bratislava, Slovakia. The study comprises two goals. The first goal of the study was to evaluate the accuracy of 3D cardiovascular printing by comparing the 3D model calliper and digital image (segmented model in .stl format) dimension measurements in all eight patients. Measurements of 4–5 dimensions (e.g. diameter of descending aorta at the level of diaphragm, right/left pulmonary artery diameter at the level of bifurcation, apex to brachiocephalic artery origin from aortic arch) individually defined for each patient were performed. Comparisons of surgical *in vivo* and 3D heart model calliper dimension measurements of specific cardiovascular dimensions were evaluated as well (e.g. diameter of transverse aortic arch, right/left pulmonary artery). Bland-Altman analysis was used for statistical analysis.

The second goal of the study was to analyse the impact of 3D heart model use for operative planning improvement in patients with complex congenital heart defects. We present the four most representative cases.

RESULTS

The Bland-Altman analysis confirmed the accuracy of 3D printing by a high correlation of dimension measurements between 3D heart models and digital images ($\pm 0.19 \pm 0.38$ mm, mean bias \pm standard deviation), as well as between *in vivo* surgical and 3D heart model at analogous anatomical locations ($\pm 0.13 \pm 0.26$ mm, mean bias \pm standard deviation) (Figs. 1, 2).

Patient 1 was a 3-day-old male neonate (weight 3.9 kg) with a clinical presentation of sinus tachycardia, dyspnoea, tachypnoea, and satO₂ difference between upper and lower extremities (88%/47%). Echocardiography visualised an interrupted aortic arch of type A. On top of that, it showed a suspicion to aortopulmonary window. The 3D heart model (Zortrax M200) derived from CT data confirmed an interrupted aortic arch type A with a 16-mm long gap between the aortic arch and the descending aorta. Also, it clearly defined the aortopulmonary window type II (communication between ascending aorta and right pulmonary artery) (Fig. 3). Because of this finding, surgical correction of this complex anatomy was performed. Ductal tissue was removed, and the posterolateral aspect of ascending aorta was end-to-side anastomosed with the descending aorta. The aortopulmonary window was transected and both orifices, aortic and right pulmonary artery, were closed by direct suture, and matrix patch plasty.

Patient 2 was a 2.5-year-old male toddler (weight 10.5 kg) with an initial echocardiographic finding of dextroversion, a double outlet right ventricle with a subaortic ventricular septal defect, and a coarctation of the aorta. Resection of the aortic coarctation with extended end-to-end anastomosis and pulmonary artery banding was performed during the neonatal period. Echocardiography and catheter angiography were performed at the age of 2.5 years. Both methods confirmed



Figure 1. Bland-Altman analysis of three-dimensional (3D) printed model measurement accuracy. Dimension measurements agreement between digital images (.stl format data derived from CT data) and 3D printed model at analogous anatomical locations. Values are expressed in mm. Central black line represents mean bias of difference. Blue lines represent upper and lower limits of agreement (LOA) \pm 1.96 standard deviation



Figure 3. Patient 1; interrupted aortic arch type A, aortopulmonary window type II. Posterior view; LPA — left pulmonary artery; RPA — right pulmonary artery; Ao asc — ascending aorta; Ao desc — descending aorta; PDA — persistently patent arterial duct; black asterisk — aortopulmonary window (connection of ascending aorta and RPA), white asterisk — interrupted aortic arch type A (missing communication between aortic arch and descending aorta)



Figure 2. Bland-Altman analysis of three-dimensional (3D) printed model measurements accuracy. Dimension measurements agreement between *in vivo* findings and 3D printed model at analogous anatomical locations. Values are expressed in mm. Central black line represents mean bias of difference. Blue lines represent upper and lower limits of agreement (LOA) \pm 1.96 standard deviation

sufficient pulmonary artery banding and a hypertrophied right ventricle, but none of them could clearly evaluate ventricular volume sufficiency, spatial relationships of both ventricles, and a spatial relationship between the ventricular septal defect and the great arteries. Consequently, a 3D heart model in two complementary parts was printed (Zortrax M200). The



Figure 4. Patient 2; double outlet right ventricle (RV), coarctation of aorta after previous aortic arch reconstruction and pulmonary artery banding. Lateral view (left side); LV — left ventricle; MPA — main pulmonary artery; AoA — aortic arch; asterisk (*) — ventricular septal defect



Figure 5. Patient 3; tetralogy of Fallot with multiple aortopulmonary collaterals, retroesophageal right subclavian artery after previous surgical central shunt (Hillel Laks) construction. Anterolateral (left side) view; RV — right ventricle; LV — left ventricle; RVOT — right ventricle outflow tract; AoA — aortic arch; Shunt — Hillel Laks shunt; LPA — left pulmonary artery; white asterisk — right pulmonary artery; black asterisk — left--sided aortopulmonary collateral



Figure 6. Patient 3; tetralogy of Fallot with multiple aortopulmonary collaterals, retroesophageal right subclavian artery after previous surgical central shunt (Hillel Laks) construction. Superoposterior view; AoA — aortic arch; Ao desc — descending aorta; lus — retroesophageal right subclavian artery ("arteria lusoria"); RPA — right pulmonary artery; white asterisk — lower right-sided aortopulmonary collateral; black asterisk — upper right-sided dual aortopulmonary collateral communicating with RPA (largest black asterisk)



Figure 7. Patient 4; tetralogy of Fallot after complete surgical correction (transannular patch including) with residual left pulmonary artery (LPA) stenosis. Anterior view; RVOT — dilated right ventricle outflow tract; LV — left ventricle; AoA — aortic arch; arrow — tortuous LPA stenosis

model revealed comparable volumes of both ventricles arranged in superior-inferior ventricular relationships (the right ventricle superiorly to the left ventricle). The apex of the heart, "formed by the left ventricle", was twisted anteriorly and to the right side. The size of the subaortic ventricular septal defect was smaller than the size of the aortic valve annulus (Fig. 4). These 3D model anatomical findings encouraged our decision-making for biventricular repair with anterosuperior ventricular septal defect enlargement, which was consequently successfully performed.

Patient 3 was a 4-month-old female (weight 5 kg) with an extreme form of tetralogy of Fallot with diminutive pulmonary artery branches (right pulmonary artery diameter 2×2 mm/left pulmonary artery diameter 1×1.5 mm), multiple aortopulmonary collaterals, and a retroesophageal right subclavian artery after previous surgical central shunt (Hillel Laks) construction. A 3D heart model based upon CT data was fabricated (Zortrax M200) 3 months after the surgery to explore the anatomy of the central shunt, the growth of pulmonary artery branches, and the anatomy of multiple aortopulmonary collaterals, as echocardiography could not clearly image these structures. The 3D model clearly depicted very complex spatial relationships among the hypoplastic right ventricle outflow tract, the central shunt, the central pulmonary arteries, and multiple aortopulmonary collaterals. It visualised an unobstructed central shunt, grown right pulmonary artery (5 \times 4.5 mm), hypoplastic left pulmonary artery (2.5 \times 1.0 mm), and origins and courses of four multiple aortopulmonary collaterals (Figs. 5, 6). Three of them had significant stenoses at their origin. One of them obviously communicated with the right pulmonary artery. The other collaterals seemed to have no communication with the central pulmonary arteries at all. Because of these findings, a catheter coil embolisation of dual aortopulmonary collateral was successfully performed. Complete correction of this complex anatomy is intended to be performed in the coming months.

Patient 4 was an 11-month-old boy (weight 7.1 kg) with tetralogy of Fallot after a previous complete surgical correction with a transannular patch. Echocardiography imaged a blurred picture of a stenotic left pulmonary artery with diminished flow. The 3D heart model (FELIX 3.0) derived from CT data showed a tortuous course of the left pulmonary artery with segmental stenosis (Fig. 7). The diameter of the stenotic segment was 3.6 mm, in comparison to the diameter of 7 mm in the distal part of the left pulmonary artery. According to these findings, Andrastent 21XL was selected for consequent catheterisational stent implantation. The successful procedure resulted in dilation of the stenotic left pulmonary artery segment to 7 mm diameter. The invasive left pulmonary artery gradient decreased from 21 to 5 mm Hg. There were no procedure-associated complications.

DISCUSSION

Three-dimensional medical model printing based on CT or MRI data has been effectively used primarily in maxillofacial and orthopaedic surgery [6, 7]. The models were described as extremely helpful for operative planning.

Three-dimensional model printing has become an established complementary imaging technique in paediatric cardiology in recent years, as well [3, 4]. The high accuracy of 3D printing in paediatric cardiology has been already acknowledged by the perfect correlation between the calliper model measurements and the patient's MRI measurements at analogous anatomical locations, with mean differences of only 0.18 \pm 0.38 mm, and 0.12 \pm 1.40 mm, respectively [3, 8]. We certified the precise accuracy of cardiovascular 3D printing in our study by a high correlation of dimension measurements between 3D heart models and digital images (+0.19 \pm 0.38 mm, mean bias \pm standard deviation), as well as between *in vivo* surgery and a 3D heart model at analogous anatomical locations (+0.13 \pm 0.26 mm, mean bias \pm standard deviation).

In comparison to the routine echocardiography, catheter angiography, MRI, or CT flat images visible on a screen, which need to be three dimensionally reconstructed in the minds of cardiac surgeons and cardiologists, printed heart models map out an exact, real 3D cardiovascular anatomy. The use of 3D models enhances physicians' understanding of the complex anatomy, so these models are valuable for perioperative planning and intraoperative orientation [9-11]. Because of this, the use of models can avoid improvisation, save intraoperative time, and improve the chances of a better outcome [2]. We experienced these invaluable benefits of 3D models in patients with a wide spectrum of complex congenital heart defects, either native or after previous surgery. Not only extracardiac (e.g. interruption of aortic arch, branch pulmonary artery abnormalities), but also intracardiac structural anomalies (e.g. spatial relationship between ventricular septal defect and great arteries in double outlet right ventricle) could be exactly evaluated. We greatly appreciated preoperative tactile opportunities to analyse the 3D heart replicas from any desired perspective long before entering the operative theatre. It allowed us to optimise surgical decision-making and better predict any perioperative complication in all patients in our study. In patient 1, with interrupted aortic arch type A, the use of a 3D model enabled us to measure the real gap between aortic arch and descending aorta, which is difficult to determine from two-dimensional (2D) echocardiographic or 2D CT pictures. It directed our preoperative decision-making from interpositum insertion to direct end-to-side anastomosis of the aortic arch with the descending aorta because the real gap distance was not as long as we assumed from echocardiography. Also, the 3D model at the same time evidently depicted an uncommon form of aortopulmonary window. Consequently, we could avoid this unexpected finding and improvisation during the surgical procedure. It could eventually shorten procedural time and patients' morbidity/mortality. In patient 2 with double outlet right ventricle, the 3D heart model, as the only imaging modality, provided us with complex information about superior-inferior arrangement and comparable capacities of ventricles, enabling double ventricle correction. In patient 3 with an extreme form of tetralogy of Fallot, in comparison to other imaging methods, the use of 3D heart model allowed us to evaluate the exact anatomy of previously constructed central shunt, the growth of pulmonary artery branches, the anatomy of multiple aortopulmonary collaterals, and complex spatial relationships of these structures.

The 3D heart models can also facilitate transcatheter interventions in patients with congenital heart defects [1, 8, 12]. We successfully used 3D models for catheter interventional planning in two patients with aortopulmonary collaterals and abnormalities of pulmonary artery branches. In comparison to limited echocardiographic imaging of aortopulmonary collaterals at 2D CT images, the 3D heart model could clearly delineate these structures in patient 3. The use of the 3D model directed us in the decision for an initial catheter coil embolisation of dual aortopulmonary collateral prior to complete correction of this complex anatomy. Similarly, in patient 4, in comparison to other imaging methods, the 3D heart model clearly depicted tortuous, segmentally stenotic left pulmonary artery eligible for stent implantation.

Limitations of the study

The limitations of the study comprise a small number of patients and a wide scale of studied congenital heart defects. The objective potential of 3D models to reduce procedural time and patients' morbidity/mortality needs to be tested in further prospective studies.

The future perspective of 3D heart model utilisation in paediatric cardiology may be associated with the improvement of bioprinting technologies.

CONCLUSIONS

Current 3D printers can produce authentic copies of patients' cardiovascular system from CT or MRI data. Patients with congenital heart defects with complex spatial anatomical relationships between cardiovascular structures should be considered candidates for this imaging modality. As we experienced in our study, the use of 3D printed cardiovascular models offering improved spatial anatomical orientation can facilitate surgical or catheter interventional procedures due to better preoperative planning and intraoperative orientation. They may eventually shorten procedural time and patients' morbidity/mortality. 3D printed models of cardiovascular anomalies can also be used as an invaluable teaching tool for paediatric cardiologists/cardiac surgeons as well as for medical students.

Conflict of interest: none declared

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Zastosowanie trójwymiarowych drukowanych modeli serca w planowaniu zabiegów u pacjentów ze złożonymi wadami serca

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Streszczenie

Wstep i cel: Celem pracy było wyjaśnienie, czy zastosowanie trójwymiarowej drukarki modelu serca może poprawić zaplanowanie zabiegów chirurgicznych bądź interwencyjnych u pacjentów ze złożonymi wrodzonymi wadami serca. W badaniu oceniano również dokładność trójwymiarowego modelu struktur sercowo-naczyniowych.

Metody: W okresie od grudnia 2014 r. do listopada 2015 r. wytworzono 8 modeli sercowo-naczyniowych, bazując na danych z tomografii komputerowej pacjentów ze złożonymi przestrzennymi relacjami anatomicznymi struktur sercowo-naczyniowych. Do oceny dokładności pomiarów trójwymiarowych wydruków z obrazami cyfrowymi oraz trójwymiarowych wydruków z tymi uzyskanymi in vivo podczas operacji zastosowano analizę Bland-Altmana. Wpływ trójwymiarowego wydrukowanego modelu serca na poprawę planowanego zabiegu oceniono u 4 najbardziej reprezentatytwnych pacjentów.

Wyniki: Analiza Bland-Altmana potwierdziła wysoką dokładność trójwymiarowego wydruku układu sercowo-naczyniowego. Każdy wydrukowany model przedstawiał poprawioną orientację przestrzenną struktur sercowo-naczyniowych.

Wnioski: Aktualnie trójwymiarowe drukarki mogą produkować autentyczne kopie systemu sercowo-naczyniowego z danych uzyskanych z tomografii komputerowej. Zastosowanie trójwymiarowych drukowanych modeli może ułatwiać zabiegi chirurgiczne lub interwencyjne u pacjentów ze złożonymi wrodzonymi wadami serca dzięki lepszemu przedzabiegowemu planowaniu oraz lepszej orientacji podczas samego zabiegu.

Słowa kluczowe: drukowanie trójwymiarowe, modele serca, wrodzone wady serca

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